

Exploring the Palliative Care Needs and Delivery of Services to Young Children with Life-Limiting Neurodevelopmental Disabilities and their Families: A Mixed Methods Study.

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Abstract

Children with life-limiting neurodevelopmental disabilities are a small but unique and important group within the paediatric population. While current policy discussions acknowledge that children with developmental disabilities are a heterogeneous group, policy prescriptions and legislation are aimed at the group as a whole which may not account for the exceptional needs of these children and their families. As a diagnostic group children with complex disabilities were identified as potentially benefiting from a comprehensive palliative care service, particularly where the child's condition involves susceptibility to health complications and the likelihood of premature death. Despite this there is relatively little known about the morbidity experienced by this particular group of children and their families, or how well current services meet their needs.

This mixed methods study explored the palliative care needs of young children with life-limiting neurodevelopmental disabilities and their families and the services that are currently available to them. A sequential explanatory mixed methods design was used to explore families experiences of providing care and engaging with services. Families participated in a postal survey (n=63) and interviews (n=12). A Delphi design, consisting of three individual interviews and a panel of 13 service providers, explored expert opinion on current services and the changes required to improve service delivery to this population of children and their families.

The findings suggest that although the children experience considerable morbidity associated with their condition, access to specialist palliative care services is not routinely required, and this group of children may be better served by improving access and frequency of the mainstream services that are currently available. The morbidity experienced by the family rivals that experienced by the child, yet families suggest that this is under acknowledged and that their engagement with services often acts to exacerbate rather than ameliorate the stress they experience. Both parents and service providers identify that current services are under-resourced and insufficient to meet the needs of this population of children. However, they do not agree issues related to how services currently function, or on the factors that would best act to improve these services.

The need to amend current practice in order to better meet the needs of this population of children and their families is evident in the findings of this study. The amendments required relate to several aspects of current practice including greater support for families caring for young children with life-limiting neurodevelopmental disabilities, improving the process of service delivery, and improving the services themselves.

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List of Acronyms

AAP	American Academy of Pediatrics
ACT	Association for Children with Life-Limiting or Terminal Conditions and their Families
CoE	Council of Europe
CHIP	Coping Health Inventory for Parents
ChIPPS	Children's International Project on Pediatric / Hospice Services
CSO	Central Statistics Office
DOH&C	Department of Health and Children
EAPC	European Association of Palliative Care
GHQ	General Health Questionnaire
HSE	Health Services Executive
ICD	International Classification of Diseases
IFS	Impact on Family Scale
IHF	Irish Hospice Foundation
MPOC	Measure of Processes of Care
MSPSS	Measure of Self Perceived Social Support
NALA	National Adult Literacy Association
NACPC	National Advisory Committee on Palliative Care
NDA	National Disability Authority
NDS	National Disability Strategy
NIDD	National Intellectual Disability Database
RCPCH	Royal College of Paediatrics and Child Health
WHO	World Health Organisation

Chapter 1: Introduction to the Study

There is nothing like looking if you want to find something.
You certainly usually find something if you look, but it is not always quite the something you were after.

J.R.R. Tolkien
The Hobbit
Harper Collins Children's Books
Re-issue Edition, 2009 p.156

1.1 Introduction

Children's palliative care attends to the physical, psychological, social and spiritual requirements of children with life-limiting and life-threatening conditions and incorporates the care of the child's family as an essential part of its remit (Sepulveda *et al*, 2002).

Although the principles of palliative care for children and adults are comparable there are many ways in which children's palliative care is unique. These include the wide spectrum of childhood conditions which may render a child in need of palliative care, the rarity of many childhood life-limiting conditions, and the fact that children requirements for palliative care is often prolonged, intermittent and at times unpredictable (Association for Children with Life-Limiting or Terminal Conditions and their Families [ACT] & Royal College of Paediatrics and Child Health [RCPCH], 1997, 2003, 2009; Department of Health and Children [DOH&C], 2001, 2005, 2010; World Health Organisation [WHO], 1998, 2002). An additional critical feature of children's palliative care is the centrality of a family-centred approach which focuses on the child and family as the unit of care, with an emphasis on the holistic care of all involved (WHO, 2002; Sepulveda *et al*, 2002; Council of Europe [COE], 2003; DOH&C, 2005, 2010).

It is within this framework of children's palliative care, as a conceptualisation of holistic, family-centred care relevant to all children with life-limiting, life-threatening and terminal conditions, that the current study is presented. The purpose of this chapter is to provide an introduction to, and set the context for the study. It begins with a definition of the terms and concepts used throughout the study, provides a general overview of the study, and presents the theoretical framework on which the study is based. The chapter concludes with an outline of the format of the remainder of the thesis.

1.2 Definition of Key Terms

In order to be clear about the focus of this study it is necessary to clearly define the terms and definitions that are used. This section introduces and defines the key terms used throughout the study.

1.2.1 Definition of Child

Irish legislation specifies a “child” to be a person under the age of 18 years other than a person who is, or has been, married (Government of Ireland, 1991). This is generally the agreed term in children’s research literature. In the context of this study the term “young child” refers to a child aged six years and under.

This demarcation point was a pragmatic rather than philosophical one. Children under six years represent a developmentally distinct population, whereas including all children up to 18 years spans the entire spectrum of childhood developmental stages. It is acknowledged that confining the study to young children may limit the scope of the findings. However, it is also proposed that confining the scope of the study to young children has distinct advantages including: children under six years represent a more homogenous sample, which eliminates the confounding variable of different developmental stages; this group of children are likely to have comparable needs and service availability. Thus confining the sample in this manner may act to increase the generalisability of the findings to similar populations. Circumscribing the population for the study was essential to identifying and accessing the population of interest which would have proved almost impossible in the absence of a rigorous database from which to draw a representative sample.

1.2.2 Definition of Palliative Care Concepts Used in the Study.

The concept of palliative care is not necessarily well understood and definitions of it vary, particularly in the context of the uncertain prognosis associated with many childhood conditions (Davies *et al*, 2008). It is essential to differentiate between earlier conceptions of palliative care as a focus on end-of-life or terminal care (WHO, 1990), and the more recent broader conceptions of palliative care which focus on holistic care throughout the illness (Sepulveda *et al*, 2002) and to clarify which focus is being used. Palliative care in this study

refers to the modern concept of palliative care as an integrated and holistic approach to the management of care for all children with life-limiting and life-threatening illnesses and their families. It is defined in accordance with the definition proposed by the ACT and RCPCH (2003:9)

“palliative care for children and young people with life limiting conditions is an active and total approach to care, embracing physical, emotional, social and spiritual elements. It focuses on enhancement of the quality of life for the child and support for the family and includes the management of distressing symptoms, provision of respite and care through death and bereavement”.

It is also necessary to acknowledge the distinction between “specialist” and “generalist” palliative care. Where reference is made to palliative care in the study it does not imply specialist palliative care services. It refers more to the palliative care philosophy or approach to care rather than to the specialist care delivered by a particular service or place of delivery. This idea that a general palliative care approach is applicable to all individuals and families facing life-threatening or life-limiting illness, in all care settings, has been widely endorsed nationally and internationally (WHO, 2002; CoE, 2003; DOH&C, 2001; 2005; 2010). Similarly in the context of this study a palliative care need is defined as a physical, psychological, social or spiritual need that is present in the context of life-limiting or life-threatening illness. It is the context of the life-limiting condition rather than the complexity of the need that designates a palliative care need. Subsequently having a palliative care need does not necessarily imply a requirement for specialist palliative care services.

1.2.3 Definition of Neurodevelopmental Disability

Neurodevelopmental disabilities comprise a diverse group of diseases with variable illness trajectories and life expectancies. There is an extensive list of neurodevelopmental conditions, but they all have dysfunction of the nervous system in common. These conditions vary along a number of dimensions including their aetiology, stability, predictability, complexity and threat to life and associated intellectual disability (Hagerman, 1999; Tager-Flusberg, 1999; Goldstein & Reynolds, 2010). Individually many of these diseases are extremely rare, but as a group they are united by their progressive nature and limited

prognosis. For such children premature death is likely although often unpredictable (Eiser, 1993).

Where this study uses the term “life-limiting neurodevelopmental disability” it refers to a condition of neurodevelopmental origin for which there is currently no cure, and which is likely to lead to the child dying prematurely. While it is acknowledged that many children with life-limiting neurodevelopmental disabilities may have long periods of relative stability interspersed with episodes of acute, potentially life-threatening illness and complications (Eiser, 1993), the overarching context of the child’s condition is one of a limited life expectancy secondary to a condition for which no curative intervention exists.

1.2.4 Definition of a Life Limiting Condition

While the terms “life-limiting” and “life-threatening” are commonly and often interchangeably used to describe the spectrum of children requiring palliative care, there has also been some debate in the literature about the ambiguity of these phrases. This study uses the term “life-limiting” in the context of the definition proposed by Sutherland *et al* (1994). Sutherland proposes that a life-limiting condition is one for which there is currently no cure and which is likely to result in the child dying prematurely, whereas a life-threatening condition is one where there is a possibility that medical intervention may prove successful. This distinction is compatible with the definition of life-limiting illnesses proposed by the ACT and RCPCH (2003:9) who define life-limiting conditions as

“those for which there is no reasonable hope of cure and from which the child or young person will eventually die”.

1.3 Background to the Study

Literature is replete with discussions of which children would benefit from access to palliative care services and under what circumstances. Although widely used, Stein *et al* (1993) have cautioned against a diagnostic approach to children’s palliative care service eligibility. They suggest that this approach relies on the presence of a specific medical diagnosis and is subject to a number of limitations, including: not every disorder to which children are subject can be included; diagnostic criteria may vary between clinicians and

clinical settings; symptoms may be present well in advance of official diagnosis; diagnostic labels do not adequately convey the extent of morbidity, and categorisation based upon medical diagnosis has an inherent bias towards children with access to medical care and services. Additionally the Children's International Project on Pediatric / Hospice Services [ChIPPS] (2001) suggests that prognosis for short-term survival should not be required as a criterion for access to children's palliative care as the illness trajectory is notoriously difficult to predict with accuracy.

The philosophical underpinning of current international paediatric palliative and supportive care models advocates that palliative and supportive care should be offered to all children with life-threatening or chronic illnesses / disabilities with complex care needs (WHO, 2002; ACT, 2003, 2009; DOH&C, 2005, 2010). It is widely accepted that an overlap exists between the care required by children with a life-limiting illness and that required by children with complex disabilities (ACT, 2003, 2009; DOH&C, 2005, 2010). Regardless of the diagnosis or nature of the condition, the challenges confronting families caring for any child with a life-limiting conditions whatever its origin are multiple, complex and specific. The needs of such children and their families require special consideration to enable the appropriate delivery of multidisciplinary care which aims to relieve suffering and improve quality of life. It is proposed that from the time of diagnosis, throughout the illness trajectory, an integrated model of palliative and curative care be adopted to allow the child and family to benefit from both philosophies of care (ChIPPS, 2001; WHO, 2002; ACT & RCPCH, 2003, 2009; DOH&C, 2005; Mack & Wolfe, 2006; Radbruch *et al*, 2007). If required, involvement in specialist palliative care services should occur early, anticipating a child's needs well in advance of deterioration and assisting with later stage symptom management and psychosocial issues (WHO, 2002; ACT & RCPCH, 2003, 2009).

1.3.1 The Irish Context

Death rates for children aged under 18 years in Ireland have declined from 5.1 per 10,000 in 2002 to 3.8 per 10,000 in 2006 with the majority of childhood deaths occurring in the period of infancy (DOH&C, 2008c). There were 1,543 childhood deaths in Ireland between 2007 and 2009 of which almost half (49%) again occurred in children under one year (Central Statistics Office [CSO], 2010). In 2008, the last year for which reliable and comprehensive

statistics are available, the International Classification of Diseases [ICD] classifications VI, XVI and XVII (WHO, 2007) accounted for 281 of the 404 deaths (69.5%) that occurred in children 0-14 years, and 272 of the 336 deaths (81%) that occurred in children under four years (CSO, 2010). It is difficult to extrapolate with any certainty from the figures that are available what proportion of these children experienced a life-limiting neurodevelopmental disability although it is probable, given the diagnostic classifications, that it would be noteworthy¹.

As a result of increasing medical sophistication many more children are now surviving with complex disabilities which require consistent and complex health care over the course of the child's life (Cohen, 1995; Alvarez, 2008). Children with life-limiting neurodevelopmental disabilities are a unique and important group within the paediatric population. As a diagnostic group they were identified as potentially benefiting from a comprehensive palliative care service, particularly where such a disability involves susceptibility to health complications and the likelihood of premature death (ACT & RCPCH, 2003, 2009; DOH&C, 2005, 2010). It is not possible to estimate the number of children dying from, or living with, life-limiting neurodevelopmental disabilities as no empirical database exists. Although there were 2,800 children (0-17 years) registered as having medical needs on the National Intellectual Disability Database [NIDD] in 2009 (Kelly *et al*, 2010), of which 40% were under 6 years of age (n=1121), the complexity of medical need these children experience cannot be identified as children are categorised only according to whether or not a medical need exists. Despite the fact that Quinn *et al* (2005) identified the urgent need for a database with agreed criteria and reporting protocols over five years ago, to date no database exists from which to assess the prevalence of this population, or indeed the prevalence of childhood life-limiting conditions in general.

Children with these conditions, particularly those who are medically fragile and have the kind of complex ongoing medical needs frequently associated with a life-limited prognosis, are a vulnerable population of children who have intensive care needs and utilise numerous health care services (Mentro, 2003). In Ireland care of these children commonly involves a variety of statutory, voluntary and charity services (DOH&C, 2010). This may increase the potential

¹ These classifications relate to diseases of the nervous system, certain conditions originating in the perinatal period, congenital malformations and chromosomal abnormalities.

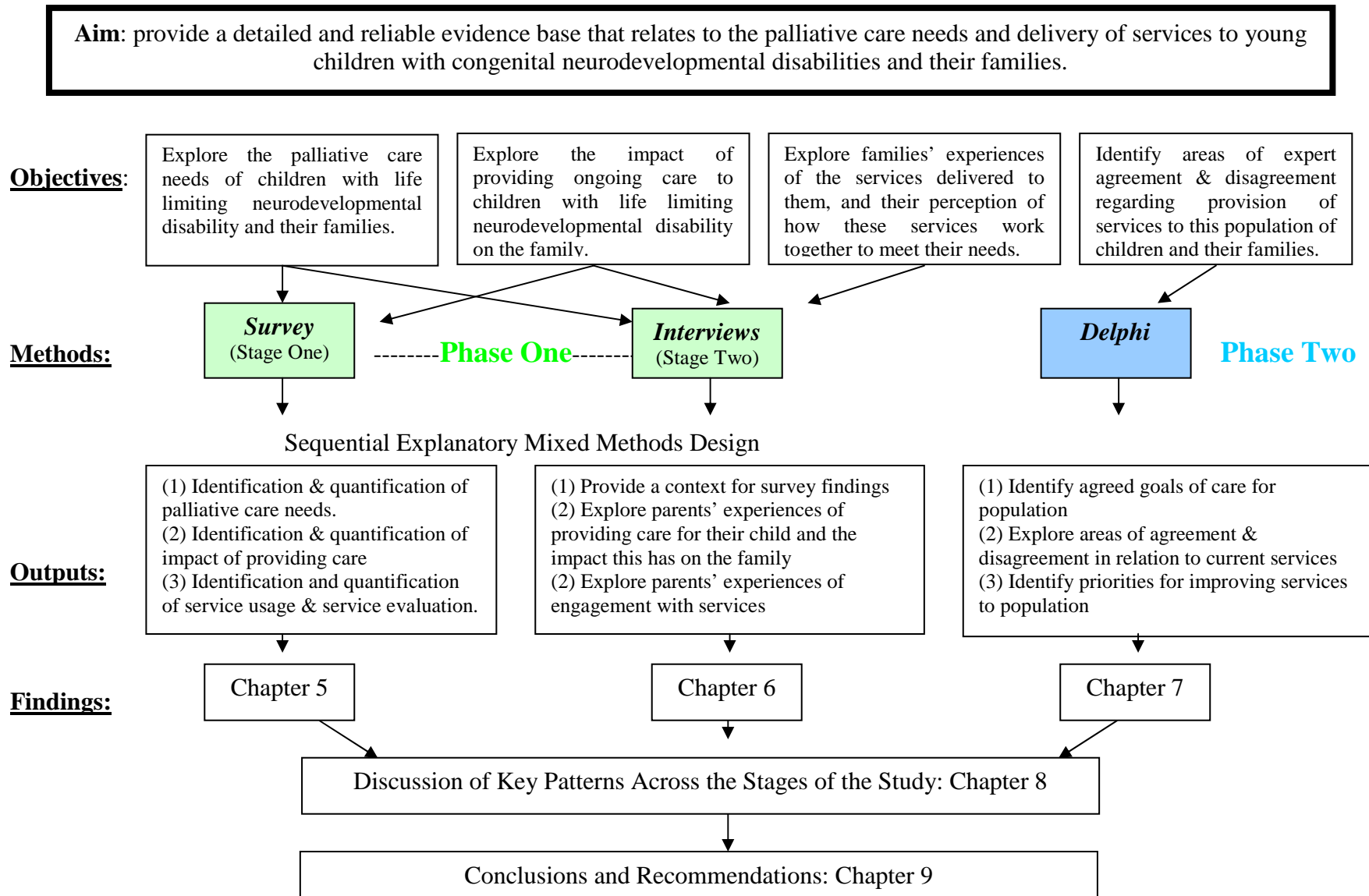
for organisational and service delivery fragmentation in the absence of collaborative working and shared expertise. Subsequently the needs of these children and their families require special consideration to enable the appropriate delivery of multidisciplinary care which aims to relieve suffering and improve quality of life. In this context it is increasingly important that there is a foundation of sound research evidence upon which to base proposed changes to policy and services. Health services research plays a key role in the approach to policymaking in any health system. It can assess the health system against defined policy objectives to ensure that the goals in providing healthcare are being met, and it can also identify shortcomings in the system. The WHO (2005) proposes that there is a strong need for evidence examining health care outcomes associated with particular policies and models of service provision. This study, which explores both the needs of children with life-limiting disabilities and their families and the services currently available to them, from the perspectives of both service users and service providers, will make an additional contribution to this research base.

1.4 Overview of the Study

This study explores the palliative care needs, and delivery of services, to young children with life-limiting neurodevelopmental disabilities and their families in Ireland. The following section presents the rationale for conducting this study including its aims and objectives.

The modes of inquiry are presented only briefly in the following section. These are discussed in detail in subsequent chapters in the thesis. Figure 1.1 maps the study's aims and objectives to the modes of inquiry and to the final outputs of the study.

Figure 1.1 *Overview of the Study*



1.4.1 Rationale for Conducting the Study

Life-limiting neurodevelopmental disabilities are relatively rare conditions, although as Zurynski *et al* (2007) demonstrate, low prevalence does not equal low impact. It is widely accepted that an overlap exists between the needs of children with life-limiting disabilities and the palliative care needs of children with a wide range of life-limiting and life-threatening illnesses (ACT& RCPCH, 2003, 2009; DOH&C, 2010). However in order to integrate and tailor palliative care services to meet these needs a comprehensive assessment of exactly what needs exist, and an evaluation of current services capacity to meet these needs is required. This evidence is not currently available, as up to this point there has been limited systematic investigation of the particular needs and experiences of this unique population of children and their families. This study addresses this deficit.

A systematic and comprehensive approach that considers the broad ranging implications of disease in all aspects of the child and their family's lives should be the standard of practice for all children with palliative care needs (Brook & Hain, 2008; ACT & RCPCH, 2009). Yet, at a general level, there is evidence that current models for the treatment of children with serious, life-limiting or terminal illnesses are inadequate, with research suggesting the provision of inadequate support services that lack integration into the mainstream of therapy (Hunt *et al*, 2003; Quinn *et al*, 2005; Steele & Davis, 2006). There is also evidence that children with disabilities have trouble in accessing the current Irish model of palliative care, and where services are available they are complex, confusing and uncoordinated (Redmond & Richardson, 2003). It would appear that many of the programmes and services available are based upon the assumptions of healthcare providers rather than the experiences of those involved, and this has inhibited the development of realistic, compassionate and integrated services to this population of children and their families.

While it is acknowledged that not all children with palliative care needs will require specialist palliative care services, the *Palliative Care Needs Assessment for Children* (DOH&C, 2005) highlights the need for a coordinated approach to age appropriate care, given in the location of choice, by healthcare professionals specifically educated and trained to care for children. In order to bridge the gap between the aspirations of NACPC (DOH&C, 2001) and the situation described in the *Palliative Care Needs Assessment for Children* (DOH&C, 2005)

the DOH&C adopted a new policy for children's palliative care *Palliative Care for Children with Life-Limiting Conditions – A National Policy* (DOH&C, 2010). This policy proposes to build upon existing frameworks and resources to deliver children's palliative care that will better meet the needs of all children with life-limiting conditions and their families. However in reality there is often a discrepancy between policy and practice, and although presenting a new and expanded vision for children's palliative care in Ireland it is probable that the resources available to implement the policy will have a significant impact upon the implementation of this new plan. In this context it is increasingly important that there is a foundation of sound research evidence upon which to base these proposed changes to policy and services. The World Health Organisation (2002, 2005) proposes that there is a strong need for evidence examining health care outcomes associated with particular policies and models of service provision. This study will make an additional contribution to this research base.

Although the *Report on a Research Study of the Palliative Care Needs of Children in Ireland* (Quinn *et al*, 2005) provides recommendations to guide the development of children's palliative care in Ireland, the authors specify that this is a generic overview rather than a model designed to meet the needs of specific groups. It is, therefore, necessary to explore the unique needs of this particular population in order to establish existing needs and to evaluate the capacity of existing services to meet these. It is important to look specifically at children with life-limiting neurodevelopmental disabilities and their families because it is possible that the genetic component involved in many developmental disabilities, as well as the complex medical regimes that often must be implemented at home, and the omnipresent threat of a life-threatening medical crisis create special circumstances for this population of children and their families. Research in this area is needed to provide the premise of fact upon which national policy must be built, and services designed and delivered. Exploring the palliative care needs of individual specific groups of children is also congruent with the general children's palliative care research agenda proposed by Quinn *et al* (2005) and Steele *et al* (2008).

1.4.2 Study Aims and Objectives

The overall intention of this study is to provide a detailed and reliable evidence base that relates to the palliative care needs and delivery of services to young children with life-limiting neurodevelopmental disabilities and their families in Ireland. The study proceeds in two phases. Phase One focuses on the perspective of the child and family and has three main objectives. These include -

- To explore the palliative care needs of children with life-limiting neurodevelopmental disability and their families.
- To explore the impact of providing ongoing care on the family.
- To explore families' experiences of the services delivered to them.

Phase Two of the study focuses on the perspectives of service providers and relates to the current provision of services to these children and their families. This second phase has one main objective which is to identify expert opinion on the issues involved in providing services to this population of children and their families.

1.4.3 Modes of Inquiry

In order to meet the objectives of the study it was necessary to employ several modes of inquiry. Consequently the study uses a mixed methods design. The first phase of the study uses an explanatory mixed methods design, and collects data using both postal surveys and interviews. The second phase of the study utilised a Delphi approach to elicit expert opinion on the issues involved in providing services to this population of children and their families. A more detailed overview of the methodology is presented in Chapter Four, with the specific method for each individual phase and stage of the study discussed in detail in Chapters Five to Seven.

1.5 The Theoretical Basis of the Study

A theoretical base was needed to underpin this study which reflected the critical concepts with which the study was concerned. This study is rooted in the concepts of family nursing and children's palliative care both of which emphasise the family as a unit of care, the delivery of family-centred care, and a focus on the family as a legitimate target for intervention.

Subsequently a theoretical framework which acknowledges the family as a central factor which can, and does, have a profound impact upon the physical and psychological health status of individual family members and the family system itself was required. This section presents the theory upon which this study is based namely Family Stress, Adaptation and Adjustment Theory (McCubbin & Patterson 1983a, 1983b; McCubbin & McCubbin, 1991, 1993; McCubbin *et al*, 1996a), and the rationale for its choice.

1.5.1 Choice of a Family Theory for the Study

There is no singular conceptual framework or theory from nursing that fully describes the relationships and dynamics of families and provides sufficiently broad knowledge on which to assess and intervene with families (Freidman *et al*, 2003). In this context theories from a variety of social science disciplines were explored as a potential basis for this study.

Amongst these were Family Systems Theory (Bowen, 1978); Ecological Systems Theory (Bronfenbrenner, 1979, 1989); and Family Stress, Adaptation and Adjustment Theory (McCubbin & Patterson 1983a, 1983b; McCubbin & McCubbin, 1991, 1993; McCubbin *et al*, 1996a).

The application of a systems perspective had particular relevance to the study in its conceptualisation of families as comprised of individual members who share a history, have some degree of emotional bonding, and develop strategies for meeting the needs of individual members and the family as a group (Bowen, 1978). However Family Systems Theory is principally related to family transactional processes, and appears to be heavily orientated towards identifying the emotional and interactional problems within the family. This study required a theory that sufficiently addressed the issue of family coping and adaptation, a pivotal feature of family-centered care when a child had a life-limiting condition or requires ongoing complex care.

Ecological Systems Theory (Bronfenbrenner, 1979, 1989) is a theory of human development which emphasises the contribution of environmental factors to human development. The potential of this theory to the current study lay in its emphasis on studying relationships amongst family subsystems. However Ecological Systems Theory has a very specific orientation towards the development of the individual rather than the family unit, explicitly

stating that contexts are always defined from the viewpoint of the developing person (Bronfenbrenner, 1989:227). Additionally, Ecological Systems Theory ultimately represents a model of normal family development and functioning, whereas this study was concerned with specific illness related factors and their consequences for the family unit.

Both of these theories had concepts relevant to this study: both acknowledge the inevitability and ubiquity of family change, and both have an interactional and holistic focus. However, this study required concepts and guidelines which were not provided by either Family Systems Theory (Bowen, 1987) or Ecological Systems Theory (Bronfenbrenner, 1989). The study is based upon the premise that in the context of childhood life-limiting neurodevelopmental disability the functioning of the family unit interacts with each individual member of the family in a discernable way which subsequently affects the health status of each individual family member and the family unit itself. In this specific context Family Systems Theory presented too broad and general an approach, with a primary focus on understanding the organisational complexity of families and the interactive patterns that guide family interactions. Conversely, Ecological Systems Theory presented too individualistic an approach, which focused more on the individual than on the processes of family coping. Consequently this study used Family Stress, Adaptation and Adjustment Theory as its theoretical foundation (McCubbin & Patterson 1983a, 1983b; McCubbin & McCubbin, 1991, 1993; McCubbin *et al*, 1996a). At a time when health policy is shifting the focus of care from hospital to community wherever possible (DOH&C, 2001a) the case for assessing and addressing the needs of families, often the primary carers, seems obvious. Although this health policy shift towards primary care underscores the family's role key in providing care and facilitating positive health outcomes, at the same time recent social policy and legislation, particularly in the context of disability services, has narrowed the focus of care with a distinct emphasis on "person-centred care".

1.5.2 Family Stress, Adaptation and Adjustment Theory

There has been a concerted effort amongst researchers over the past decades to account for the observed differences among families in their coping with, and adaptation to, stressful situations, and a substantial amount of cross-fertilisation amongst the theories developed. An example of this is Family Stress, Adaptation and Adjustment Theory (McCubbin & Patterson

1983a, 1983b; McCubbin & McCubbin, 1991, 1993; McCubbin *et al*, 1996a) which borrows heavily from Ruben Hill's seminal "ABCX Family Crisis Model" (Hill, 1949; 1958), and draws heavily on family systems and family development theory. According to McCubbin and Patterson (1983a) this new theory established a link between Hill's original Family Stress Theory (1949; 1958) and the subsequent physiological and psychological theories of stress developed by Lazarus (1966) and Selye (1974).

Family Stress, Adaptation and Adjustment Theory is based upon the Double ABCX Model of Stress and Adaptation (McCubbin & Patterson 1983a; 1983b), which utilised Hill's original key concepts of a stressor event; the family's crisis meeting resources and the meaning the family makes of the event to explain a family's response to stress. According to Hill (1949, 1958) a family's susceptibility to crisis is determined by the balance between these key factors. While Hill's original model focused primarily on pre-crisis variables that account for differences in families' capabilities and adaptation, McCubbin & Patterson (1983b) proposed that assessing families' post-crisis behaviour required a more dynamic model that focused on family efforts over time. The Double ABCX Model of Family Adaptation (McCubbin & Patterson 1983a, 1983b) expanded upon and supplemented Hill's original key concepts in an effort to describe the additional life stressors which shape the course of family adaptation [Appendix A]. These post-crisis variables included: the critical psychological, intra-family and social resources families acquire and employ to manage crisis situations over time; the changes in definition and meaning families develop in an effort to make sense of their situation; the coping strategies families employ; and the range of outcomes of these family effort.

Stress is not a pejorative term within Family Stress, Adaptation and Adjustment Theory, but neutrally described as a life event or transition impacting upon the family unit which produces, or has the potential to produce, change in the family social system (McCubbin and Patterson (1983a:1983b). In the context of this study this stressor is represented in the child's condition and the attendant care work involved which has long been identified as a chronic family stress in the context of childhood disability (Ray & Ritchie, 1993; Dyson, 1997; Keating, 1997; Kearney & Griffin, 2001; Datta *et al*, 2002; Redmond & Richardson, 2003),

and similarly identified in the context of childhood life-limiting conditions (O'Brien, 2001; Steele & Davies, 2006; Katz, 2002)

Rather than Hill's concept of a single stressor event, McCubbin & Patterson (1983a, 1983b) propose that families experience an accumulation or pile-up of demands over time. They refer to these as "aA" factors within theory model, suggesting that the demands may originate from a variety of sources including: individual family members, the family system, or the community of which the family is a part. In the context of this study this accumulation of demands represents the ongoing difficulties associated with the child's health status, the difficulties encountered in managing the child's care, the impact of care provision on other family members and the family unit, and the social world outside the family with which they must engage.

Family adaptive resources are termed "bB" factors in McCubbin & Patterson's model (1983a, 1983b). These are defined as "part of the family's capabilities for meeting demands and needs" (McCubbin & Patterson, 1983b:14). They suggest two distinct types of adaptive resources: existing resources, which are already part of the family repertoire and serve to minimise the impact of the initial stressor; and expanded family resources, which are new resources which are strengthened or developed in response to the new demands. "cC" factors refer to the meaning that the family gives to the total crisis situation (McCubbin & Patterson, 1983b). This is influenced by perceptions of the stressor believed to have caused the crisis as well as the added stressors, the old and new resources available to the family, and estimates of what needs to be done to bring the family back into a state of equilibrium or balance.

Families attempts to redefine the crisis situation generally involve efforts to clarify the issues, hardships and tasks involved in the crisis in an attempt to render them more manageable and responsive to problem solving efforts; decrease the intensity of the emotional burden associated with the crisis situation: encourage the family to carry on with its fundamental tasks of promoting the social and emotional development of its members. Thus, according to the Double ABCX model while resources, perception and behavioural responses interact as a family attempts to achieve a balance in family functioning, coping, which has both cognitive and behavioural components, becomes a bridging concept between bB and cC factors.

In the Double ABCX model the “xX” factor refers to family adaptation and balancing. McCubbin & Patterson (1983b, 1983b) suggest that three elements need to be considered in family adaptation: the individual family member; the family system; and the community of which the family members and family unit are a part. A balance needs to be achieved between the individual member and the family unit, and between the family unit and the community. Each of these elements is characterised by both demands and capabilities. According to the model “family adaptation is achieved through reciprocal relationships where the demand of these units is met by the capabilities of another so as to achieve a “balance” simultaneously at two primary levels of interaction” (McCubbin & Patterson, 1983b:18). Family adaptation is the central concept, and outcome measure, of Family Stress, Adaptation and Adjustment Theory. The concept is used to describe a continuum of outcomes which reflect efforts to achieve a balanced “fit” at the levels of member-to-family, and family-to-community. This adaptation continuum ranges from “Bonadaptation” (positive adaptation which signifies positive outcomes) to “Maladaptation” (which signifies negative outcomes or the emergence of a crisis situation) (McCubbin & Patterson 1983b:13).

1.5.3 Relevance of Family Stress, Adaptation and Adjustment Theory to the Study

From the perspective of Family Stress, Adaptation and Adjustment Theory the child and the family are inextricably linked: the health of one affecting the health of the other through dynamic and reciprocal interaction effects. The theory provides a relevant and sound theoretical framework for this study in its conceptualisation of the family as a dynamic, developmental and functional unit and a legitimate focus of care which is compatible with the philosophical roots of this study. The theory is widely used in family nursing representing a strengths based approach to families with a focus on adaptability (Garmezy, 1991). In addition, its shifting of the outcome variable from family crisis to adaptation reflects the evolution of a more strengths and resiliency orientation of family stress and disability researchers (Freidman *et al*, 2003; Darbyshire & Jackson, 2004). It also plays a vital and positive role in explaining support and in-home care to promote the wellbeing of family members who may be affected by illness (De Marco *et al*, 2000).

Using Family Stress, Adaptation and Adjustment Theory and the Double ABCX Model Patterson & McCubbin (1983a, 1983b) identify eight potential sources of family stress in the context of childhood chronic illness. These include strained family relationships; modifications in families' activities and goals; the burden of increased tasks and time commitments; increased financial burden; social isolation; the need for housing adaptation or modification; medical concerns (related to obtaining competent medical care; understanding, clarifying and verifying medical information; the ability to follow through with prescribed home treatment; how to help the child endure or minimize pain and other symptoms; and worry and uncertainty regarding the child's prognosis); differences in the child's school or social experiences; and grieving associated with developmental delays or abnormalities, restricted life opportunities for the child, and, for some illnesses, anticipation of an early or painful death. These sources of family stress have since been reiterated in research literature related to children with chronic, life-limiting conditions and complex disability. Additionally the theory has been shown to be a useful theoretical basis for assessing families in primary care (Tomlinson, 1986; Robinson, 1997), the context in which most care is provided for children with life-limiting neurodevelopmental disabilities. The theory has previously been applied as a conceptual framework for exploring childhood life threatening illness (Brody & Simmons, 2007) and various aspects of childhood disability (Giallo & Gavidia-Payne, 2006; Van Riper, 2007; Levine, 2009), and is particularly relevant to the practice of family nursing because it emanates from a professional practice heritage rather than an academic discipline.

The previous sections of this chapter have presented the background to this study, and the concepts and definitions used throughout. The final section presents the structure of the remainder of this thesis.

1.6 Format of Thesis

The remainder of this thesis proceeds as follows.

Chapter Two provides the policy and service contexts in which care is currently provided to children with life-limiting neurodevelopmental disabilities and their families. Policy and service contexts are explored in advance of the research literature as this study was primarily driven by policy and practice and subsequently informed by theory and research.

Chapter Three provides a critical review of the research literature related to the subject of this study. Gaps in the current corpus of knowledge are identified, and these are related to the aims of the study. The literature review is structured in accordance with the study's theoretical framework.

Chapter Four presents a detailed overview of the current study. It includes an account of the overall mixed methods design used, and the research questions to be addressed. The impact of the theoretical framework on the concepts explored in the study, and the manner in which this exploration was operationalised, is discussed. Finally the ethical principles on which the study is based are presented and their application is discussed.

Chapters Five, Six and Seven provide details of the specific methods used in each stage of the study namely survey, interview and Delphi methods. The design, method of data collection and analysis, and the findings of each stage are presented independently with preliminary discussion.

The findings from both phases of the study are integrated in Chapter Eight with key patterns across the stages identified and linked back to the research questions. In this chapter key patterns are discussed in detail and placed in the context of previous research. The chapter concludes with a discussion of the overall strengths and limitations of the study.

Chapter Nine draws conclusions about these findings and offers recommendations based upon these. Future directions for research are also proposed.

1.7 Conclusion

This chapter has provided an introduction to the current study including a definition of the key terms used throughout. The background and rationale for conducting the study has been presented, and the aims and objectives of the study have been described. The theoretical basis on which the study proceeds is presented. Finally the structure and content of the remainder of the thesis has been presented.

Chapter 2: Policy and Service Contexts

Health systems also reflect their societies.
Their development needs to be driven, not only by outcomes, but by shared values.
They all share the same overall goals: health gain, fairness, and responsiveness.

W.H.O. (2005) *Strengthened Health Systems*.
W.H.O. Regional Office for Europe, Copenhagen. (p.6)

2.1 Introduction

This chapter explores the policy and service milieu in which care is delivered to children with life-limiting neurodevelopmental disabilities and their families in Ireland. In the context of this population of children the links between health policy and disability policy are strong. In addition it is impossible to ignore the international dimension of evolving concepts of children's palliative care, and the impact this has had on the development of national health policy for these children and their families. The chapter begins with a discussion of some of the general issues involved in children's palliative care, in particular the challenges and complexity of defining the population from a service perspective. The case for adopting a palliative care approach to children with life-limiting neurodevelopmental disabilities and their families is presented. Finally the structure and policy context of current and proposed Irish children's palliative care services will be discussed. There will also be a brief discussion of pertinent disability policy and services. However, the emphasis will be on elucidating the convergence and divergence between the philosophies underpinning the palliative care and disability policy areas.

2.2 Children's Palliative Care

Increasing medical and technological sophistication has increased the longevity of many children who would previously have died from conditions associated with prematurity, congenital abnormalities, and other degenerative conditions (Goldman, 1998, 2003). Consequently the number of children living with chronic conditions for which there is no cure, and which require complex and ongoing health care over the course of the child's life, continues to increase (Brook & Hain, 2008). In this context children's palliative care may be

needed for a wide range of illnesses, which differ from adult diseases, and many of which are rare and familial (Goldman, 2003). Indeed one of the unique features of children's palliative care is the wide spectrum of conditions which may render a child in need of such care with many children requiring palliative care having life-limiting conditions, as opposed to advanced terminal conditions, with which they may survive for many years (Goldman, 2003; Brook & Hain; 2008; Hain & Wallace, 2008). A significant proportion of these children will have long-term needs and many live with severe disability, in this context while a palliative care approach is appropriate these children may not necessarily require specialist palliative care services (DOH&C, 2001, 2005, 2010). Palliative care for children and young people with life-limiting conditions has been defined as

“an active and total approach to care, which embraces physical, emotional, social and spiritual elements. It focuses on enhancement of quality of life for the child / young person and support for the family. It includes the management of distressing symptoms, provision of short breaks and care through death and bereavement” (ACT & RCPCH, 2009:7).

Although children's palliative care evolved from the specialty of paediatrics its model of care has evolved from that of traditional adult services. In common with adult palliative care services it struggles with definitional and operational difficulties, not least that of clearly defining its target population. Finding succinct language to describe the breadth and complexity of children's palliative care continues to pose an ongoing challenge, and the identification of those children who would best benefit from access to palliative care services, and the manner in which these children would best be served, remains somewhat elusive. In an attempt to address the issue of which children would benefit from access to palliative care the English based children's charity ACT have specified four categories of childhood life-limiting conditions for which palliative care is appropriate and beneficial (ACT & RCPCH, 1997, 2003, 2009). This categorisation provided the first common definition of children who would potentially benefit from palliative care and has been widely referred to in children's palliative care literature [Figure 2.1]. In an Irish context, the categorisation was used as a framework by Quinn *et al* (2005) as the basis of a review of the palliative care needs of Irish children. It has also been adopted by the DOH&C (2010) as the framework for the national policy *Palliative Care for Children with Life-Limiting Conditions in Ireland*.

Figure 2.1 *Categories of Childhood Life Limiting Conditions for Which Palliative Care may be Required*

<p>Group 1 Life-threatening conditions for which curative treatment may be feasible but can fail. Palliative care may be necessary during periods of prognostic uncertainty and when treatment fails. Examples: cancer irreversible organ failures of heart, liver and kidneys.</p>	<p>Group 3 Progressive conditions without curative treatment options, where treatment is exclusively palliative and may commonly extend over many years. Examples: Batten’s disease, mucopolysaccharidosis.</p>
<p>Group 2 Conditions where there may be long periods of intensive treatment aimed at prolonging life and allowing participation in normal childhood activities, but premature death is still possible. Examples: cystic fibrosis, muscular dystrophy.</p>	<p>Group 4 Conditions with severe neurological disability which may cause weakness and susceptibility to health complications, and may deteriorate unpredictably, but are not usually considered progressive. Examples: severe multiple disabilities, such as following brain or spinal cord injuries, including some children with severe cerebral palsy.</p>

However while ACT’s categorisation proves useful in terms of demarcating the remit of children’s palliative care certain operational difficulties persist, and its application to clinical practice is not always straightforward. Nicholl (2007) found that the classification is not universally used amongst Irish children’s nurses, and suggests that possibly because of its diagnostic focus, it may not accurately reflect the complex nature of the nursing and care needs of these children. In addition there is the challenge posed by the ambiguity of the phrases “life-limiting” and “life-threatening” which was highlighted in the previous chapter. While these terms are commonly, and often interchangeably, used to describe the spectrum of children requiring palliative care, research literature has been confounded by the semantics of whether conditions are life-threatening or life limiting. Sutherland *et al* (1994) propose a straightforward distinction suggesting that a life-threatening condition is one where there is a possibility that medical intervention might prove successful, whereas life limiting conditions are those for which there is currently no cure and which are likely to result in the child dying prematurely. However, in reality, children may often oscillate between these two definitions. This is especially true in an acute exacerbation of a chronic condition where the illness trajectory is almost impossible to predict with any degree of accuracy and it can be difficult to draw a distinction between what constitutes a life prolonging intervention and what is symptom management (ChIPPS, 2001).

2.3 Extending Palliative Care to Children with Life-Limiting Neurodevelopmental Disability

Children with life-limiting neurodevelopmental disabilities represent a group of children who have been identified as potentially benefiting from access to palliative care (ACT & RCPCH, 1997, 2003, 2009). As a group they are united by their ultimately limited prognosis, and for many of these children premature death is likely, although often unpredictable (Eiser, 1993; Goldman, 1998, 2003). Some children with life-limiting neurodevelopmental disability will have long periods of relatively good health while others may require palliative support at an earlier stage of their disease. Although neurodevelopmental disabilities vary along several dimensions there is also some commonality, particularly in terms of the impact on, and coping by the family. It is acknowledged, both nationally and internationally, that there is an overlap between some of the needs of children with life-limiting conditions and the care that children with disabilities will require, particularly where such disabilities are likely to lead to premature death (Quinn *et al*, 2005; Craft & Kileen, 2007; DOH&C, 2010).

The number of children surviving with complex disabilities which require consistent and complex health care over the course of the child's life is likely to continue to increase as a result of increasing medical sophistication (Cohen, 1995; Alvarez, 2008). However the consequences of increased longevity can be oversimplified, for example, by equating technologically prolonged survival with indefinitely prolonged high quality of life. When this oversimplified view is embraced, unrealistic optimism may relegate palliative care as irrelevant or misguided for these children, despite the fact that the prognosis for such conditions remains ultimately fatal (Birnkrant & Noritz, 2008). The sequelae of survival in these situations are often complex (Hain & Wallace, 2008), and it has been argued that it is entirely appropriate to call the care required by such children "palliative care" since for many of these children cure of the underlying condition was never possible (Goldman, 2003; Lenton *et al*, 2004; Carter & Leveton, 2004). For these children medical care frequently consists largely or exclusively of life-extending therapy and of comfort therapy that improves the child's quality of life (Goldman, 2003; Lenton *et al*, 2004), and Carter and Leveton (2004) suggest that access to palliative care for such children can frequently be hampered by the artificial boundaries that often separate palliative care from other forms of medical care, especially curative care, in children with complex health care needs.

2.4 Background to Palliative Care Policy in Ireland

The past fifteen years have seen the publication of a significant number of national reports, plans and strategies which have directly impacted on, or have significant implications for, the development of general palliative care services in Ireland (DOH&C, 1994, 1996, 2001, 2001a, 2004c, 2005, 2010). The publication of the seminal *Report of the National Advisory Committee on Palliative Care* (DOH&C, 2001) has possibly been the most significant and far reaching of these. This report made several recommendations regarding the funding and delivery of specialist palliative care services based upon the principles of equity, accessibility, and the adoption of a coherent and comprehensive national policy framework for palliative care. The National Advisory Committee on Palliative Care [NACPC] (DOH&C, 2001:32) differentiate three distinct levels of palliative care and proposed that palliative care services should be structured to accommodate these levels of ascending specialisation [Figure 2.2].

Figure 2.2 *Levels of Palliative Care*

Level 1	Palliative Care Approach	Involves engagement in the principles of palliative care, where required, by all health professionals
Level 2	General Palliative Care	Viewed as the intermediate level of expertise, with engagement in palliative care being part of the health professional's caring role but not defining it.
Level 3	Specialist Palliative Care	Whose practice involves palliative care as its core activity and is directed towards caring for patients with complex and demanding palliative care needs.

Although primarily focused on adult palliative care, the NACPC also made recommendations for children's palliative care. Amongst these were that "palliative care for children is best provided at home with the family closely supported by the primary care team and a specialist palliative care team where available" (p.48). The report also emphasises the importance of collaboration and coordination of services for children and families emphasising the requirement for a broad multidisciplinary approach.

The vision for palliative care services presented by the NACPC was adopted as official government policy in 2001. Despite this, and the substantial effort and significant progress made since its publication, the *Baseline Study on the Provision of Hospice / Specialist Palliative Care Services in Ireland* (IHF, 2006) confirmed marked regional disparities in

current government spending on palliative care services in all care settings. Similarly, in the specific context of children's palliative care, the *Children's Palliative Care Needs Assessment* (DOH&C, 2005) identified several continuing problems in relation to children's palliative care services in Ireland. These included issues of access and equity, unmet palliative care needs, fragmentation and lack of service coordination, and a lack of education and training for healthcare professionals dealing with children with life-limiting conditions.

In response to this needs assessment, and informed by the NACPC report (DOH&C, 2001), the DOH&C (2010) published its policy framework for children's palliative care entitled *Palliative Care for Children with Life-Limiting Conditions – A National Policy*. Underpinned by the principles of inclusiveness, partnership, comprehensiveness and flexibility the policy is aimed at addressing the issues identified in the needs assessment in order to construct a palliative care service that is responsive to the needs of children and their families. The policy document explicitly states that

“palliative care services for children should be accessible, equitable, flexible and appropriate, and should meet the needs of any child with a life-limiting condition and their family” (p.6).

Although *Palliative Care for Children with Life-Limiting Conditions in Ireland – A National Policy* has been publicly endorsed by the Minister for Children (DOH&C, 2010c), neither the policy itself, nor the Minister's speech, alludes to any timeframe for its implementation.

2.5 International Models of Children's Palliative Care

There is no “right way” to provide children's palliative care and each programme or model of care should be contextually suited to meet the resources and cultural expectations within its own locale (Fowler-Kerry, 2006). However the principles for establishing programmes and models of children's palliative care are well established (American Academy of Pediatrics [AAP], 2000; ACT & RCPCH, 2003, 2009; Radbruch & Payne, 2009). In their report on the development of children's palliative care programmes and services the AAP (2000) recommended the following principles by applied: the introduction of palliative care at the diagnosis of a life threatening illness, the education of health care providers in pediatric palliative care principles, and need for increased research support. The AAP recommends

that, regardless of the specific model used, programmes should ensure that child and family preferences are incorporated into treatment plans, care is seamless across all settings, there is continuity and consistency of caregivers of multiple disciplines, and caregivers skilled in all aspects of pediatric palliative care are always available. Similar principles for developing children's palliative care services have been proposed in the United Kingdom by ACT and RCPCH (2003, 2009). ACT suggests that truly integrated children's palliative care services should be based upon a system where universal, targeted and specialist provision work together in a coordinated way that enables both local accessibility for support and management of everyday problems, and access to first class specialist services for the management and care of more complex symptoms. In a review of paediatric palliative care in Europe, Dangel (2002) suggests that home care is the most suitable and recommended model of palliative care for children. Similarly, the more recent recommendations from the European Association of Palliative Care [EAPC] also propose that in children's palliative care the family home should remain the centre of caring wherever possible, with every family having access to a multi-disciplinary, holistic paediatric palliative care team at home (Radbruch & Payne, 2009).

At an international level these principles have been applied in various models of delivery. In the USA, Friert (2009) describes four basic models of service provision of children's palliative care which he suggests can be combined to provide a continuum of care for children with life-limiting illness. These include hospital based programmes, free-standing paediatric hospice facilities, hospice based programmes, and community agency or long-term care based programmes. Where previously access to children's palliative care in the USA has been hampered by strict rules and regulations, since September 2010, under the *Patient Protection and Affordable Care Act*, the requirement for children to have forgone curative treatment in order to be eligible for hospice care has been dropped by health insurers (National Hospice and Palliative Care Organisation, 2010).

While many countries are actively engaged in the development of children's palliative care services, the United Kingdom had been at the forefront in providing services specifically designed to attend to the needs of children with life-threatening, life-limiting and terminal conditions (Quinn *et al*, 2005; DOH&C, 2010). In the U.K. children's palliative care is

provided through statutory services such as health, social services and education and voluntary services such as hospices and other charitable organisations. Craft and Kileen (2007) suggest that the voluntary sector, and the children's hospice movement in particular, have been leaders in the development of children's palliative care services and have remained a vital partner in service delivery and innovation ever since. In a review of palliative care services to children and young people in England Craft and Kileen (2007) report that children's palliative care is delivered through a combination of specialist and generalist medical care, community children's palliative care nursing teams, charities and voluntary service providers, social care providers, and hospice care. Two forms of hospice care are described: residential children's hospices which provide a range of specialist respite care; and hospice-at-home care, which consists of a multidisciplinary team who visits the child in their own home and provides the medical and nursing care they would receive if they were an in-patient in a hospice. It is this "hospice-at-home" model that is the proposed model for the development of Irish children's palliative care services (DOH&C, 2010).

Models of children's palliative care may be applied singularly or in combination, and while there is little evidence evaluating the efficacy of children's service models, adult service evaluation suggest that there is little robust evidence to support the view that one model of service delivery is superior to another (Sailsbury *et al*, 1999; Garcia Perez *et al*, 2009). It is acknowledged that, in the context of children's palliative care, there is no single care deliver model that will work for every child or in every setting (ChIPPS, 2001).

2.6 Irish Model of Children's Palliative Care

As previously described many international models of children's palliative care provision exist and the manner of the organisation of palliative care services is heterogeneous. Currently, while Irish adult specialist palliative care services are relatively well established (Ling & O'Siorain, 2005; O'Reilly, 2005; IHF, 2006), in contrast children's palliative care services are not yet well developed, and palliative care for children has not been well integrated into the existing national guidelines (Redmond & Richardson, 2003; Quinn *et al*, 2005). Attempts are being made to address this situation and, based upon the recommendations of the *Palliative Care Needs Assessment for Children* (DOH&C, 2005), the

first specialist children's palliative care consultants has recently been appointed by the DOH&C². Additionally the first Irish children's hospice, Laura Lynn House, has recently been completed (although the palliative care needs assessment for children found mixed reviews amongst respondents and did not identify the provision of this service as a priority (Quinn *et al*, 2005). This new children's hospice has eight beds and opened its doors on September 17th. 2011. The project has been funded entirely from charity donations, and commitment has yet to be received from the DOH&C with regards to the ongoing administration of the service.

Presently most children with life-limiting conditions in Ireland are cared for in their homes and the services provided are generally community based (O'Reilly, 2005; DOH&C, 2010). The majority of the day-to-day medical care of these children is the responsibility of the primary care team (General Practitioner and Public Health Nurse), with support from voluntary bodies, and in some cases from adult palliative care services where needed (DOH&C, 2005). Despite the fact that it had previously been recommended that medical and nursing care should be provided by paediatric trained staff with the close support of a specialist palliative care team (DOH&C, 2001), it has been identified that many of these health professionals lack the necessary expertise to care for children and their families (Quinn *et al*, 2005). The overall care of the child is usually shared with a regional hospital or one of the country's three tertiary paediatric hospitals (O'Reilly, 2005).

In its report *A Palliative Care Needs Assessment for Children* (DOH&C, 2005) the Department of Health and Children identified an urgent need to develop specialist children's palliative care posts to spearhead the establishment of services and education (medical and nursing initially), yet such posts have been slow to materialise. Similarly the identified need for a 'key worker' for each child and family to co-ordinate and implement a plan of care, liaise between the family and all services, and provide families with a single resource to aid in the day-to-day management of their child's illness has largely been unaddressed. Up to this point the promise of eight specialist "outreach" posts in children's palliative care for children with life-limiting conditions has resulted in three permanent clinical nurse specialist posts nationally.

² This post is being funded by the Irish Hospice Foundation (a charity organisation) for a period of five years (Finn, 2010).

Most recently the policy document *Palliative Care for Children with Life-Limiting Conditions in Ireland –A National Policy* (DOH&C, 2010), proposes a strategy for the further development of children’s palliative care services. Built upon the findings of the children’s palliative care needs assessment (DOH&C, 2005) among its recommendations is that the Health Services Executive [HSE] should develop an implementation plan, and that a consultant paediatrician with a special interest in children’s palliative care should be appointed along with regionally based children’s outreach nurses (although this recommendation was originally proposed by Quinn *et al* five years earlier). It also proposed that a national database of children with life-limiting conditions be developed and a National Development Committee for Children’s Palliative Care be established. The policy recommends that children’s palliative care services should be advanced within the framework of the “Primary Care Strategy” (DOH&C, 2001a) with its emphasis on the development of community services. It is proposed that, in accordance with best practice, appropriate structures are put in place to enable children with life-limiting conditions gain access to care at home. These structures are specified as: the support of a key worker; provision of care as required; timely provision of aids and appliances; and a range of flexible respite care including in-home respite. With a focus firmly on family-centred care, *Palliative Care for Children with Life-Limiting Conditions in Ireland –A National Policy* proposes the “Hospice-at-Home” model for the delivery of children’s palliative care services in Ireland (DOH&C, 2010).

While this national policy is to be welcomed as a specific framework for the delivery of palliative care to children with life-limiting conditions it also contains some caveats. Explicitly stated is that children’s palliative care services must be developed “within the context of services and resources that are available” suggesting that “within the context of current financial constraints it is necessary to have a phased implementation of this policy” (p.22). In addition, despite the difficulties of defining the terminal phase in these children, the policy also proposes that the structures are put in place “with priority given to those approaching the end of life” (p.13). This focus on terminal and end-of-life care may inhibit easy access to palliative support for the many children whose illness trajectory is uncertain, and in whom it is difficult to predict the timing of the terminal phase.

2.7 Disability Policy in Ireland

The current service context and future policy direction of children's palliative care for children with life-limiting conditions has been discussed. However children with life-limiting neurodevelopmental disabilities lie at the intersection of health and disability policy, and the provision of care to these children and their families is influenced by both.

At an international level the past two decades have seen the models that inform policy prescriptions for disabled people undergo radical changes based upon the move to a social model of disability which was critical of the traditional medical approach in favour of a more socio-political approach (Barnes, 2005). The sociology of disability, underpinned by the social model, has been one of the most significant intellectual and political developments of the recent past (Chappell, 1998). It has transformed the meaning of disability at a personal, intellectual and political level and, in focusing on the ways in which disability is socially constructed, has shifted debates about disability from biomedically dominated agendas to discourses about politics and citizenship (Oliver, 1990). The social model of disability separates disability from impairment and attributes disability to the dominant socio-cultural environment (Corker, 1998), and in this respect it embodies the theoretical basis for emancipatory politics (Barnes, 2005). Gabel and Peters (2004) suggest that evidence for the adoption and influence of the social model abounds in international declarations and conventions, in national legislation, in global expansion of Community-Based Rehabilitation Programmes, in the growing number of Disability Studies degrees in universities, and in the push for inclusive education at primary and secondary school levels.

Within this broad political context Irish disability policy and services have been transformed in the past decade by a combination of new legislation and the publication of several national reports, plans and strategies (Government of Ireland, 1999; 2004; 2004b, 2004c; Department of Justice, Equality & Law Reform, 2004; DOH&C, 2005, 2007). Principle amongst these in terms of overall impact has been the publication of the *National Disability Strategy* (Department of Justice, Equality & Law Reform, 2004) and the *Disability Act 2005* (Government of Ireland, 2005). As a consequence of the *National Disability Authority Act* [NDA] (Government of Ireland, 1999) the National Disability Strategy [NDS] was launched in 2004.

The overall aim of the NDS is the promotion and support of equal participation of people with disabilities in society. The NDA required that disability policy and programmes relevant to the lifecycle framework for people with disabilities be progressed through the NDS, with particular expression being provided through sectoral plans and other relevant mechanisms. As an adjunct to this the Comhairle (Amendment) Bill 2004 (Government of Ireland, 2004b) extended the statutory basis for advocacy through the introduction of a personal advocacy service for people with disabilities, and plans for a specially appointed Director of this service. The National Disability Strategy makes no distinction between physical and intellectual disability, nor does it differentiate between different levels of intellectual disability. Instead the strategy adopts a generic position whereby they define disability as

“a substantial restriction in the capacity of the person to carry on a profession, business or occupation in the State or to participate in social or cultural life in the State by reason of an enduring physical, sensory, mental health or intellectual impairment”. (Government of Ireland, 2005:6)

Part 2 of the NDA commenced for children under the age of 5 years with effect from 01st. June 2007. This part of the NDA established a system for the assessment of individual health service needs occasioned by the disability and, where appropriate, education needs for persons with disabilities over age 18 years.

The publication of the National Disability Strategy marked a very significant milestone for the disability sector in Ireland, and signaled the move from a medically driven model to a social model of care for all individuals with disabilities regardless of the nature of the disability. Underpinning the NDS are the principles of equity, inclusion and a person-centered approach to policy and service planning. An individually focused and person-centered philosophy is implicit in both the *Education for Persons with Special Educational Needs Act* (Government of Ireland, 2004c:7) which proposes that assessment must be “in accordance with the best interests of the child” and the Disability Act which proposes that service plans “be appropriate to meet the needs of the applicant concerned” (Government of Ireland, 2005:11). In addition the DOH&C (2007:8) states explicitly that the assessment of need is “person-centered at all stages”.

2.8 Intersection of Children's Palliative Care and Disability Policy

At a general level there are some commonalities between the basic tenets of palliative care and disability services. Both are built around the fulcrum of a fundamental respect for an individual's personhood, and both advance goals of respect and dignity and promotion of quality of life. However the manner in which these goals are pursued, the philosophical underpinning of the services, and the overall orientation of services, are significantly different.

Irish legislation and policy has resulted in the development of disability services which are focused specifically upon the individual needs of the child, i.e. "person-centred". Conversely children's palliative care services are orientated towards the needs of the child and the family as a single unit or "family-centred". This may be a subtle but important distinction in the context that there is an overlap between some of the needs of children with life-limiting illnesses and the care that children with disabilities will require (DOH&C, 2010). Such differing philosophies and agendas may result in conflicting goals of care, children and families falling between services, or in failure to meet the needs of the family as a unit where the child has a combination of disability, complex medical needs and a life-limited prognosis. This has been suggested by previous Irish research which propose that current models for the treatment of these children are inadequate, lacking adequate support services and integration into the mainstream of therapy (Redmond & Richardson, 2003; Quinn *et al*, 2005). It would appear that the provision of effective palliative care for these children and their families is contingent upon overcoming the barriers between different philosophies of care, and the sharing of expertise between all services involved in the child's care so that collaborative management and integration of all services can be achieved.

2.9 Conclusion

As a result of the complex nature of their conditions, children with life-limiting neurodevelopmental disability and their families occupy a space at the intersection of health and social care. The services that these children and their families receive are heavily influenced by the policy context of both philosophies of care. This chapter has provided an overview of the Irish policy and service contexts in which care is currently delivered to this

population of children and their families. The broader international debates and perspectives which have impacted on the development on these national policies and service initiatives have been discussed.

Chapter 3: Research Literature Review

I have yet to see any problem, however complicated, which, when you looked at it the right way, did not become still more complicated.

Paul Alderson
New Scientist 1969 p.638

3.1 Introduction

This chapter presents a critical review of the research literature related to the subject area of the study. The literature review is structured in accordance with the study's underlying theoretical framework, Family Stress, Adaptation and Adjustment Theory (McCubbin & Patterson 1983a, 1983b; McCubbin & McCubbin, 1993). In keeping with the study's theoretical underpinning the birth of a child with a life-limiting neurodevelopmental disability is considered the crisis event for the family. The research literature review focuses on the post-crisis variables identified in the theoretical framework, with a particular emphasis on the demand factors associated with crisis event since this was the main focus of this study. The review is presented in three main sections: the first section explores literature related to the pile-up of family demands; section two explores families' resistance resources with a particular focus on formal support services as extra-familial resources; while the final section explores literature related to family perception and the meaning attributed to the situation. The literature is critiqued at each stage and its relevance to the specific population of this study is discussed, finally gaps and limitations in the literature are identified and these are related to the aims of the study.

Before the search strategy used to retrieve literature is described there is a brief discussion of some issues that arose relative to the use of terminology as it applies to the study's population. The complexity and ambiguity surrounding the terminology applied to children with life-limiting neurodevelopmental disability necessitated the application of a relatively broad range of search terms when reviewing the literature.

3.2 Terminology and the Selection of Studies for the Current Review

Developing systems to serve children with life-limiting neurodevelopmental disabilities and their families needs a clear definition of the population to be served. However this is not always clear in the research literature which is replete with a wide range of terminology used in reference to children with life-limiting conditions. Not only are various, and often undefined, terms used to describe these children, but the terms themselves are often used interchangeably and appear to change regularly. This issue has been a topic of debate in the literature, particularly in relation to the benefits and drawbacks of collective versus specific terminology. It has been suggested that broad and inclusive collective terminology (for example “life-limiting illness” and “complex needs”) benefits the relatively small number of children affected by specific conditions but who together often face similar hurdles and restrictions in their lives (Eiser, 1993, Hornby, 1995). Similarly Closs (1999) suggests that diagnostic approaches may risk pathologising and marginalising children if their use is extended beyond the medical field, and Stein and Silver (2002) report substantial overlap in the numbers and characteristics of the children to whom different non-categorical conceptual definitions apply.

However, it also been demonstrated that such collective categorisation includes a heterogeneous group of children, whose health conditions manifest along a continuum characterised by increasing complexity, levels and types of limitations, the presence of co-morbidities, and the need for specific types of health services (Newacheck & Taylor, 1992; Stein *et al*, 1993; Stein, Westbrook, & Bauman, 1997; Newacheck & Halfon, 1998; Neff *et al*, 2002). Sutherland *et al* (1994) and Nicholl (2007), whilst not advocating the use of diagnostic labelling, propose that the range of undefined terminology in the literature may result in confusion and poor understanding by both providers and users of health services. Similarly Bramlett *et al* (2009) suggests that the heterogeneity of collective terminology can present a challenge for developing systems of care that are appropriately responsive to the need of individual groups of children.

The lack of agreement regarding the definitions and terminology associated with childhood life-limiting conditions makes identifying any specific sub-population of children a

complicated task, and posed a considerable challenge in conducting the literature review for this study. Although individual diagnostic labels were not used to recruit participants to the study, the general term “life-limiting neurodevelopmental disability” does represent a loosely circumscribed diagnostic group of children, albeit a diagnostic group that is not specifically identified in the research literature. This makes confining and reporting literature related only to these children as a distinct, individual and specific group almost impossible. Nevertheless when examining the inclusion criteria or the description of the samples used in many research studies related to life-limiting childhood conditions it is apparent that these children frequently form a small subsample of many of the samples used. Consequently the literature for this study was reviewed in the context of the inclusion criteria applied to participants. The sample of children in this study had two primary characteristics: a neurodevelopmental disability, and a limited life expectancy as a consequence of this complex disability (this was the primary ground on which participants were recruited). However, because of the difficulties encountered with service providers when the term “life-limited” was used in conjunction with these children, it was required that this be evidenced using specific criteria. These inclusion criteria are described in detail in Chapter Four, they broadly included: having a known diagnosis associated with a life-limited prognosis, having ongoing complex medical needs such as a dependence on medical intervention and associated technology, or the presence of complex symptoms that were difficult to control.

In this context all of the inclusion criteria for the study were considered when conducting the research literature review. Literature related to children with general life-limiting illnesses was considered relevant on the basis that the substantive issues involved in the care of the care of life-limited children should not be drastically different regardless of diagnostic categorisation, and the fact that it is widely accepted that an overlap exists between the care required by children with a life-limiting illness and that required by children with life-limiting disabilities (DOH&C, 2005, 2009). There is no universally agreed definition of the term “complex health need” subsequently the term has also been variously used in research literature to describe different populations (Nolan *et al*, 2005; Condliffe, 2006; Watson, Abbott & Townsley, 2006). Stalker *et al* (2003) suggest that children who are described as having complex and continuing healthcare needs are often dependent on some form of medical technology, or require regular and unpredictable drug administration, or have

difficult to manage symptoms. This term is a term frequently applied to the children who this study investigated; subsequently this literature was also reviewed in the context of this study. Other studies identify children who are dependent on technology as a distinct population consequently this literature was also included as it too formed part of the inclusion criteria for the study. Research literature related to neurodevelopmental disability was also reviewed as this was the second element of the child's diagnosis.

The basic assumption underlying the research literature review was that, whatever the origin of the child's condition, and regardless of the diagnosis or nature of this condition, the challenges confronting children with any life-limiting conditions and their families are multiple, complex, specific and often overlapping. Davis and Brosco (2007) suggest that all definitions of child health and categorisation of childhood illnesses are essential contextual and that flexibility is critical for researchers with a specific goal and limited access to certain samples or populations. The issue, they suggest, is that researchers be clear about which classification scheme was used, and the implications of this for the study findings. Although a relatively broad range of research literature was reviewed for this study all of the studies reviewed involved children with life-limiting disabilities or complex medical needs amongst the samples. However the inclusion of such a broad range of literature makes it difficult to be certain about how the research findings relate specifically to the children and families with which this study is concerned. An added difficulty with the research literature was the inclusion of a broad range of children's ages in the majority of study samples. Again, this raises a question about the generalisability of the findings to any specific age group of children as the degree to which findings are influenced by the inclusion of children of all developmental ages cannot be known.

3.3 Search Strategy

A systematic search of the published literature was undertaken in advance of conducting this study with periodic reassessment over the full course of the study from 2008 to 2011. The search used electronic databases including Cumulative Index of Nursing and Allied Health Literature [CINAHL], ChildData, Cochrane Library, Health Source Nursing, Medline, Proquest, Blackwell, PsychArticles, ScienceDirect. InterNurse and Synergie. The database Dissertation Abstracts was used to identify unpublished theses in the area, and major texts

and policy documents from the Irish government, Council of Europe and World Health Organisation were searched. Literature from voluntary organisations including Association for Children with Life-Limiting or Terminal Conditions and their Families [ACT], Irish Association for Palliative Care [IAPC], Irish Hospice Foundation [IHF] were located and reviewed as were official Government publications relevant palliative care, children with life-limiting illnesses, and children with disabilities. Search engines such as Google Scholar and PaedPalLit were also used.

Searches were made using appropriate database subject headings. In association with the terms “child” and “children”, the initial keywords “life-limiting”, “life-threatening” and “neurodevelopmental disability” were used. In the context of the difficulties associated with terminology other key related terms such as “complex (healthcare) need”, “technology dependence”, and “medically fragile” were also used in association with the keywords. Searches made use of broader terms, narrower terms, Boolean operators (and, or) and truncated terms. Available scholarly book literature was also reviewed. No date parameters were applied to the literature at the outset although where possible the focus was on literature published from 2005-2011. The search was confined to English language material only.

3.4 Prevalence of Childhood Life-Limiting Conditions and Disability

There is an increasing number of children living with chronic conditions, for which there is no cure, and which require complex and ongoing health care over the course of the child’s life (Goldman 1998, 2003). Similarly more children with life-limiting complex disabilities and technology dependency are living for longer because there is more that can, and is expected, to be done for them (Craft, 2004). Beyond the fact that this is a growing population there is relatively little research on the morbidity experienced by this group of children, and robust empirical data on the prevalence of children with life-limiting conditions is largely lacking.

The epidemiology of childhood deaths has radically altered over the last century (Feudtner, 2000; Himmelstein *et al*, 2004). In the developed world a substantial number of children die from chronic life-limiting disorders and conditions, frequently present and diagnosed at birth

(Feudtner, 2000; Davis & Higginson, 2002; Rallinson & Moules, 2004). International estimates for the number of children living with life-limiting conditions are variable. Roughly similar figures are reported for the USA, 10 per 10,000 children (Institute of Medicine, 2002; Hynson *et al*, 2003) and the U.K, 12 per 10,000 children (Lenton *et al*, 2001), although Hain (2005) reports a higher figure of 3.75 / 10,000 for Wales. In an Irish context, Quinn *et al* (2005) used a combination of the prevalence figures from the United Kingdom (ACT, 2003), the ICD-10 classification of diseases (WHO, 2007) combined with population data from the CSO, to propose an estimated national prevalence of 1,369 children living with life-limiting conditions, and a mean mortality rate of 370 per annum, in Ireland between 1996-2001. However they also suggest that the figures be treated with caution as they are based upon the assumption that the U.K and Irish prevalence figures are similar. The latest information provided by the Central Statistics Office for 2002-2004 shows that there has been a decrease in the number of childhood deaths from all causes, whilst the number of deaths from life-limiting conditions remains almost static at 3.5 per 10,000. Information also indicates the majority of childhood deaths continue to occur in the first year of life (DOH&C, 2008c). However, in reality, the absence of agreed definitions and accurate statistics means that the exact number of children, or the magnitude of their need, remains at best a rough guess.

There were 2,800 children (0-17 years) registered as having medical needs on the National Intellectual Disability Database [NIDD] in 2009 (Kelly, Craig & Kelly, 2010), of which 40% were under 6 years of age (n=1121). It is not possible to identify the complexity of medical need these children experience since children are categorised only according to whether or not a medical need exists. One of the limitations of the NIDD database is its failure to account for the tremendous variability in terms of the type and severity of medical need and the subsequent requirement for, and use of, health care services. However, it is likely that children with life-limiting disability are likely to form only a small, but significant, subset of the population of children registered on the NIDD database. Although there is no robust data upon which to base this claim it is consistent with the findings of Quinn *et al* (2005) who reported very limited primary care experience (General Practitioners and Public Health Nurses) of children with severe neurological disability.

3.5 Demands and Risk Factors for Family Adjustment: aA Factors

There is a considerable body of knowledge supporting the notion that caregiver stress is a common phenomenon among families caring for children with life-limiting conditions and complex needs (Thyen *et al*, 1998; Baxter, Cummins, & Yiolitis, 2000; Weiss, 2002; Saloviita *et al*, 2003; Singer, 2006; Reichman, Coman, & Noonan, 2008; Miodrag & Hodapp, 2010). According to Family Stress, Adaptation and Adjustment Theory (McCubbin & Patterson 1983a, 1983b; McCubbin & McCubbin, 1993) family outcomes related to the impact of this stress are the result of a multitude interconnected factors associated with the stressor and its related hardships. This section of the research literature review explores issues associated with the risk factors for family adjustment and adaptation, namely the “pile-up of demands” associated with providing ongoing care. These include: the morbidity experienced by the child and the nature of the work involved in providing ongoing care, which provides an indication of the stressor and its severity; and the family impact of care provision which relates to the associated hardships cause by the crisis.

3.5.1 Morbidity Experienced by Children with Life-Limiting Conditions

Possibly as a consequence of the relatively small number, and wide variety of life-limiting illnesses, there is relatively little research literature exploring the specific morbidity experienced by children with life-limiting conditions. Despite a comprehensive assessment of the palliative care needs of children with life-limiting conditions in Ireland Quinn *et al* (2005) did not elicit data on the specific health status or problems experienced by the children involved in their study, nor did Redmond and Richardson’s earlier Irish study (2003) which explored young children with severe / profound and life-limiting complex disabilities.

The only study to specifically and directly assess the physical morbidity experienced by children with life-limiting conditions was conducted by Lenton *et al* in 2001. These researchers used a cross-sectional survey to determine prevalence and associated morbidity of non-malignant childhood life-limiting conditions, in children aged 0 – 19 years, in one health district in the United Kingdom. Considerable morbidity amongst the sample of 93 children was reported including mobility, sensory, cognitive and communication difficulties,

feeding and sleep disorders. Additional 59% of the children in the sample experienced pain or severe discomfort that was not being effectively controlled. The degree of disability and range of problems exhibited by the children indicated high levels of medical, nursing and psychological needs. When contrasted with children with malignant life-threatening illness, Lenton *et al* (2001) concluded that children with non-malignant life-limiting conditions generally have a poorer outlook, both living longer and with greater levels of disability. This study was the first and indeed only attempt to quantify the health problems associated with childhood life-limiting illness. The researcher administered survey had many strengths not least the fact that the inclusion criteria were clearly defined and the probability of death related to the child's condition was explicitly stated. Perhaps the major limitation of the study, in terms of practice and policy, was its failure to comprehensively address the issue of symptom management in the sample of children. The presence of a symptom or problem does not necessarily imply that it is not well controlled or managed. Its relevance to this study included the incorporation of children with life-limiting complex disabilities amongst the sample. Although the age range of children was much broader than in this study this is unlikely to affect symptomatology. However, Lenton *et al's* study (2001) excluded seven children who died before the assessment, and eight children whose families were considered too distressed during the terminal phase of their child's illness. The study therefore described the chronic symptoms experienced by children with life-limiting conditions rather than those experienced in the terminal stage of the child's condition, and it is not possible to ascertain whether symptom profile escalated, or changed significantly, in the terminal stage of the child's condition.

In the absence of direct empirical evidence about the morbidity and needs of children with life-limiting conditions, Donnelly *et al* (2005) used a concept mapping exercise with 50 health professionals from a variety of practice disciplines in the USA to develop a seven-cluster concept map of children's needs. This was not confined to physical morbidity and the final model included clusters of needs related to pain, decision making, medical system access and quality, dignity and respect, family-oriented care, spirituality, and psychosocial issues. However, all members of this study were health-professionals associated with the provision of palliative care, or advisors to palliative care committees. It is therefore probable that their perception was based upon their professional experience i.e. children involved with

palliative care services. This does not represent the situation for the children in this study who are involved with a range of statutory and voluntary services. Indeed Donnelly *et al* (2005) represent only a particular professional perspective related to a particular group of children. It is difficult to extrapolate the findings to children and families not in receipt of palliative care service since there were no children or families involved in the development of this model of need.

Despite consistent calls for access to palliative care services (WHO, 2002; CoE, 2003; ACT & RCPCH, 2003, 2003b, 2009), research literature related to the actual morbidity experienced by children with life-limiting conditions is largely lacking (in contrast with the morbidity experienced by the families of these children). However, despite this lack of direct investigation, there are studies of the nature of the work involved in caring for these children which may provide a more oblique indicator of the children's morbidity.

3.5.2 The Nature and Management of the Child's Care

Although the number of children with life-limiting and complex disabilities is small relative to the number of children with general disabilities, the medical, physical, social and emotional care of these children involves proportionally more time, physical and financial resources (Campbell, 2007). The overall survival rate of children with life-limiting conditions and complex life-limiting disabilities is increasing (DOH&C, 2005; 2009) and this is in keeping with international trends and projections (Alvarez, 2008). In an Irish context most of these children are cared for at home and the services provided are community based (O'Reilly, 2005; DOH&C, 2010). Caring at home for a child with any chronic condition requires both cognitive and physical work on the part of parents, who face many adaptive tasks in the course of their ongoing care work. These include: expertise in managing symptoms, adapting their environment, adjusting their life-style, meeting the needs of other family members, coping with ongoing stress and periodic crises, and establishing a support system (Canam, 1993; Balling & McCubbin, 2001).

Children with life-limiting, life-threatening conditions or complex disability often require specialised medical and technical care that would more usually be delivered by trained health professionals (Redmond & Richardson, 2003; Quinn *et al*, 2005). The physical workload can

be relentless, with parents frequently required to assume the responsibilities of a professional nurse but without entitlement to a day off (Glendinning & Kirk, 2000, Wang & Bernard, 2004). In addition these children are more vulnerable to acute illnesses, often requiring frequent hospitalisations for both acute illness and exacerbation of their chronic condition (Burke *et al*, 1989, Canam, 1993; Berman *et al*, 2005). Children with non-malignant life-limiting illnesses are also subject to frequent oscillations in their condition making the trajectory of their illness difficult to predict with any certainty (Brook & Hain, 2008). Despite this, there has been relatively little research undertaken in terms of quantifying the work or time involved in caring for a child with a life-limiting condition or complex medical needs at home. Chase and Rogers (2001) suggest this is a consequence of the perception that women's care work continues to be constructed as essentially a private issue located within the domestic sphere of the home.

Two studies have attempted to quantify the care and time costs of caring for a severely disabled child, both of which were undertaken in the U.K. Roberts and Lawton (2001) conducted a review and analysis of the quantitative records of 40,500 families of severely disabled children in an attempt describe the specific care and extra attention parents provide. The data on 4,500 infants (<1 yr.) and 35,000 children (1-16 yr.) were reviewed. In the 1-16 yr. age group, on average each child needed extra care with six activities. In the infant group, while all infants obviously required total care, severely disabled infants required additional care. The majority of infants had extra care needs associated with every daily activity. Requirements for extra care around feeding and during the night were particularly common. Qualitative data was particularly helpful in identifying the nature and cause of the extra care required: for example extra care associated with feeding could be due to the need for extra feeds, tube feeds, and swallowing difficulties which could be particularly time-consuming and entail a risk of choking. Other parents reported that their infant could not be left alone, even for a few minutes, because of the danger of fits or choking. Virtually all needed extra care during the night because of the potential for medical crises.

The groups were combined to estimate that, on average, each severely disabled child needed extra care in six areas of daily life when compared to a non-disabled child. Nine out of ten infants and four out of ten children had recurrent medical crises. The sample also included

660 children who were technology dependent. These children had additional care needs beyond those of other children in the sample including extra general care needs, overnight care needs, and more medical crises. On average these children needed extra help with seven areas of daily life and additional specialised care associated with their medical technology. The generalisability of this study to young children with life-limiting neurodevelopmental disabilities is limited by the fact that the sample is not well described, with the term “severely disabled” applied as a catch-all for a variety of chronic physical and intellectual disabilities and life-limiting conditions including cancer, heart disease and lung disease. The analysis of infants separate from other children provides some insight into the difficulties experienced by younger children, although why only two age classifications were considered relevant (infants and all other children) is not clear, and makes it impossible to establish whether the child’s needs changed with developmental stage. Despite these limitations, Roberts and Lawton’s study (2001) included children with life-limiting neurodevelopmental disabilities amongst its sample. In addition the segregation of infants from older children, and the different patterns of need described, suggests that younger children may have additional, or more complex, needs.

A second study, also undertaken in the U.K, focused on the time costs of caring for a severely disabled child. Curran *et al* (2001) conducted a comparative analysis of parents caring for children with severe disabilities (n=16) compared with parents caring for normally developing children (n=31). This study reported that items of personal care were significantly different in the group of children with disabilities than in the non-disabled group. In addition, at least one third of the families of a child with a disability had to attend to the child’s care at least once during the night. In contrast to Roberts and Lawton’s study (2001) the higher frequency of personal care items did not decrease with increasing age but was associated with level of disability as measured by the Functional Disability Score although the difference in classification of disability between the two studies may account for this finding. Curran *et al*’s study (2001) did not make reference to the time parents of children with severe disabilities spend attending appointments, accessing and dealing with services, or providing therapeutic interventions for their child. This may imply that the findings in fact underestimate the actual time costs of care provision for these parents. The

fact that the sample of children with severe disabilities ranged in age from 3-17 years limits the generalisability of the findings to the current study.

The nature of the work involved in caring for a child with a life-limiting condition has received considerable attention from a qualitative perspective, where studies describe consistent patterns and trends. Similar to quantitative investigations these studies suggest that the work involved in caring for such children is considerable. They also report that while parents often needed help to alleviate the impact of their caregiving this was not always available in a useful manner.

Parents in Steele and Davies's (2006) grounded theory study of eight families caring for children with a neurodegenerative life-threatening illness, describe care that is constant and unrelenting. The children in this study required care 24 hours a day, seven days a week. Steele (2005) suggests that care becomes increasingly complex as the child's condition changes or deteriorates. In qualitative interviews with seven mothers of children with a wide range of disabilities Green (2007a) reports that mothers describe their experience of providing care as both time consuming and physically exhausting. Redmond and Richardson (2003) describe how the nineteen mothers in their study had acquired quite sophisticated skills in the areas of nursing and physiotherapy in order to care for their young severely / profoundly and life-limited disabled child. Similar findings were reported by Kirk and Glendinning (2002, 2004) who reported that, in the day-to-day management of care, the parents of the 24 technology dependent children in their study performed highly technical clinical procedures which in hospital would be considered the domain of professionals. Kirk and Glendinning (2002) propose that the level, complexity and intensity of the nursing care which these parents provide is distinctive, and sets them apart from all other parents. In addition to the actual hands-on care provided, the constant vigilance required by the child is exhausting for parents and siblings (O'Brien, 2001, Steele & Davies, 2006; Nicholl, 2008). Both Nicholl (2008) and Manaseri (2008) highlight an additional feature of the work and expertise involved in caring for a child with complex medical needs. Nicholl's (2008) phenomenological investigation of 17 mothers describes the "pre-emptive" aspect of caring for children with complex needs, whereby not only do mothers need to be able to respond to the child's current needs, but they need to be able to anticipate and act to prevent any

potential crises. Additionally, in a grounded theory study of 18 mothers Manaseri (2008) describes how mothers not only need to understand and provide the complex medical routines and procedures involved in the care of their child, but also need to be able to temper this knowledge and adapt it to what is “normal” for their own child.

These studies are subject to the limitations of any small scale study in terms of sample size, with participants purposefully selected through service providers or support groups. This may suggest that the findings represent a particular population of parents which may not be generalisable to all families in similar situations. As the studies were descriptive in nature they did not attempt to provide any measure of the care work undertaken by parents. However, the findings of qualitative explorations of the nature of the care provided to children with life-limiting conditions or complex medical needs are consistent in describing a considerable burden of work for parents.

In summary, findings from quantitative assessments of the practical burden involved in caring for a child with a severe disability are consistent in terms of reporting the significant extra care needs that such children require, and the fact that this also involved night-time care. However, although Roberts and Lawton’s (2001) findings suggest that this care requirement diminishes with age, conversely Curran *et al* (2001) suggest that this is not the case, and that the higher frequency of care items did not decrease with age but was instead associated with the child’s level of disability. It is likely that the different definitions of disability between the two studies accounted for this discrepancy. Both of these accounts inform the current study in the context that both included amongst their samples children with severe neurodevelopmental disabilities, some of whom were dependent on various forms of medical technology. They did not however focus specifically on children with life-limiting neurodevelopmental disabilities, nor were these studies focused on a particular age group of children. In this context the degree to which these findings can be generalised to the population of children in this study is unclear.

With the exception of Redmond and Richardson’s study (2003) qualitative investigations of the care required by children with life-limiting conditions were not specifically focused on children with neurodevelopmental disabilities, but involved a wide variety of children with

life-limiting conditions, complex disabilities, or children dependent on medical technology. In addition, the inclusion of a broad range of age groups in many samples potentially limits the extent to which the findings can be applied to any specific age group of children. However overall, studies exploring the nature of the work involved in caring for a child with a life-limiting condition or severe and complex neurodevelopmental disability are consistent, and suggest that the complex nature of the work involved in caring for these children requires skills of vigilant assessing, interpreting, and acting on the child's conditions all in the context of knowing that a miscalculation could have significant consequences for the child. The consistency of these finding across many studies, both quantitative and qualitative, adds weight to the body of evidence suggesting that the care these children require is persistent, complex, highly skilled, and highly burdensome. In addition, the consistency of the findings, across many samples and regardless of the nature of the child's condition, suggests that similar difficulties are encountered across diagnostic categorisations.

3.5.3 *Balancing and Structuring Family Life*

Since most of the care provided for children with life-limiting conditions or complex life-limiting disability occurs in the child's home, caring for such children clearly has implications beyond the child and the care provided. Parents are faced with emotional, physical, social and financial impacts on their lives, and expend considerable psychological and physical energy as they attend to the cognitive and practical aspects of caregiving. Caregiving demands affect all facets of family life (Alexander *et al*, 2002), which is characterised by frequent change and unpredictability (O'Brien, 2001). Using a cross-sectional survey and follow up interviews with three culturally different groups of self-selected mothers McConkey *et al* (2008) report increased levels of poor maternal physical and psychological health, increased child-related stress particularly associated with pessimism about the future, and poorer family function that are consistent across cultures.

Care for a child with a life-limiting or complex medical condition or who is assisted by medical technology can be considered a chronic stressor for the family (Thyen *et al*, 1998; Miodrag & Hodapp, 2010). Parents struggle with emotional strain, the physical and psychological dependence of the child, the impact on family relationship, and the feeling that there was "no free choice" in the matter (Carnevale *et al*, 2006; Nicholl, 2008). Similarly

families of children with developmental disability are at risk of having higher levels of stress in comparison to families of typically developing children (Singer, 2006; Weiss, 2002), with Baxter *et al* (2000) reporting that the stress experienced by families with a child with a developmental disability was twice that experienced by families without a disabled child. The impact on parents of having a child with a complex disability is pervasive and multidimensional and similarly can impact on all members of the family unit and affect all aspects of family functioning (Reichman *et al*, 2008). The goals for any family living with a child with a complex or life-limiting condition or dependent on medical technology are targeted to achieve some degree of stability so that optimum child and family development can be maintained, and to gain control over family life (O'Brien, 2001; Alexander *et al*, 2002; Steele & Davies, 2006; Carnevale *et al*, 2006), however achieving this stability and managing daily family life in this context represents a constant challenge.

In a qualitative study of 15 purposefully selected families of children, aged 3- 12 years who were technology dependent, O'Brien (2001) described family life as characterised by a constantly changing balance and the need for the family to continuously restructure. This was associated with areas of child health status, schedules and routines and family roles and responsibilities. O'Brien (2001:13) describes this potential for frequent and unpredictable change as "living in a house of cards", suggesting that this state of constant unpredictability and uncertainty and limited parental control contributes to families perceptions of the fragility and instability of their lives. Areas of challenge, growth and change identified by families in this study included: the dimensions of making sense of family life, managing daily life with technology dependence, maintaining a functioning family, and negotiating with outside entities. The families in this study were purposefully selected through health care agencies, social care agencies and parent support groups, and thus represented a specific section of parents. Although all parents had been caring for their child at home for a period of at least one year, the range of technologies on which the child was dependent was vast, ranging from gastrostomy feeds to ventilator dependence. The nature of the study makes it impossible to determine whether the experience differed amongst families' related to the complexity of the child's technology dependence. However the study was relevant to the population in the current study in the context that technology dependence was identified as an inclusion criteria in the current study.

Similar findings were reported by Alexander *et al* (2002) in a grounded theory study of 5 families caring for a child who was dependent on technology. In contrast with O'Brien's (2001) study, all of the children in this study were ventilator dependent at home. The children in this study ranged in age from 8 – 16 years. Alexander *et al* (2002:7) report a central theme of “struggling daily” as characteristic of the experience of family life in the context of child technology dependence as mothers described struggling to constantly re-organise in the face of constant unpredictability and uncertainty. Three distinct dimensions of the struggle were identified: getting over the hump, described as the most difficult and overwhelming process and families learn to care for the child and manage their family situation; starting to breathe, a period of relative stability when families have managed to successfully negotiate the challenges; and dealing with a subsequent event, which resulted in the family the family having to manage the situation with renewed intensity. Alexander *et al* (2002) report that families experiences appear to be profoundly influenced by three intervening conditions: the accessibility and availability of formal resources, the involvement of family and friends, and the family's socioeconomic status. However this claim is made without discussion in the main body of the research and in the absence of supporting interview text from the couples involved in the study. Although the relevance of Alexander *et al*'s (2002) study to the target population for this study lies in the technology dependence of the children, beyond this fact the samples are significantly different in both age range and type of technologies used. In this respect it is not clear well the findings of the study can be generalised to the population of interest in the current study.

Steele and Davies (2006) report findings from a grounded theory study of eight families caring at home for a child with a neurodegenerative life-limiting condition. Unlike the previous two studies there is no description of the children in this study beyond their diagnostic category. These authors propose a model of “navigating uncharted territory” to describe the journeys that families caring for their life-limited child undertake (Steele & Davies, 2006:576). The model describes four main dimensions within this process: entering unfamiliar territory, shifting priorities, creating meaning, and holding the fort. The authors describe how all aspects of the process are pervaded by the significant physical, cognitive and emotional work that parents undertake in the care of their child. However unlike O'Brien (2001) and Alexander *et al* (2002), Steele and Davies (2006) also report that parents

described positive aspects of caring for their child. Despite this observation there is little comment or explanations of this aspect of care in the study. Since there is little description of the children in this study's sample the degree to which it can inform the current study is not clear. Its relevance lies in the fact that all of the children in the study had a diagnosed life-limiting illness. Similar findings are reported by Carnevale *et al* (2006) in the context of children assisted by technology. In an interpretative qualitative study of 12 families these authors describe the process of living with a ventilator-dependent child as "daily living with distress and enrichment" (p.48). Like the parents in Steele and Davies study (2006) the families in this study also described deep enrichment and rewarding experiences that they could not imagine living without. However this occurs against a backdrop of overwhelming and stressful parental responsibility and the feeling that there was really no "free choice" in the matter.

Rodriguez and King (2009) used a hermeneutic phenomenological approach to explore ten mothers lived experience of providing care for a child with a life-limiting condition in the United Kingdom, describing six continuing constituents of the mothers' lived experience: inner drive, feeling responsible, psychological effects, threatened self-image, social withdrawal, and fear of reaching the bottom line. Similar to previous studies mothers in this study described lives where they never stopped and were continually running from one task to the next and felt responsible for everyone and everything. Rodriguez and King (2009) describe several aspects of psychological distress in the mothers in this study, reporting frustration, anxiety, insomnia, and feelings of lack of control. They also describe how mothers had their energy drained by the constant struggle with the health system to get what they needed and suffered threatened self-image and social withdrawal as a consequence of caring for their child. The final theme described in this study, fear of reaching the bottom line, described how in various ways all parents, at different times, had almost reached the point of emotional breakdown in situations where inner pain and fight had gotten so severe. Unlike the studies by Steele and Davies (2006) and Carnevale *et al* (2006) there is little account in this study of a positive dimension to this situation beyond the comment that mothers appeared to achieve a sense of satisfaction from their inner drive to provide care. This study used the small sample congruent with the research approach; however the mothers were selected through their acquaintance with the author in the context of a larger evaluation

study. It is not clear why these particular mothers were chosen over others involved in the larger study, and the impact of this has implications for the findings of the study.

In summary, although there were variations in the study designs and approaches used, studies exploring the nature of family life when a child has a life-limiting condition or technology dependence suggest that supporting and caring for the child at home makes the management and the maintenance of family life a constant challenge. Children with complex medical needs require an extraordinary quantity and quality of care, which demands careful orchestration and places considerable demands on parents. Studies suggest that family life is characterised by uncertainty and constant struggle (O'Brien, 2001; Alexander *et al*, 2002; Steele & Davies, 2006; Carnevale *et al*, 2006; Nicholl, 2008; Rodriguez & King, 2009) and feelings of both lack of control and lack of choice (Nicholl, 2008; Rodriguez & King, 2009), although some studies report a positive dimension to family life (Steele & Davies, 2006; Carnevale *et al*, 2006). In keeping with the nature of the phenomena, studies exploring the experience of family life are small scale studies using purposefully selected samples, and as such are subject to questions about the scope and transferability of the findings. In addition the age range of the children in the studies was broad, which make it difficult to account for the impact of the different developmental stages of the child and limits the generalisability of the findings to the population in the current study. In the case of O'Brien's study (2001) a considerable range of technologies were included which would suggest considerable variations in the children's conditions. However, despite the various samples and limitations, findings are consistent across qualitative investigations suggesting that, while there may be differences in how the process unfolds for families, families share a common experience regardless of the nature of the child's condition or type of technology used. Similar to studies describing the nature of the work involved in caring for a child with a life-limiting condition or disability, the inclusion of a broad range of age groups in many samples potentially limits the extent to which the findings can be applied to the specific age group in this study, and the nature of the children's conditions may limit the generalisability of the findings to children specifically with life-limiting neurodevelopmental disabilities.

3.5.4 Impact on the Family Home

Caring for a child with complex health care needs impact not only on the nature of family life and the people involved in the provision of care, but also on the very environment in which this care is delivered. Children with complex health care needs typically require technical and / or medical equipment in the home, both because of their need for intensive ongoing care and to compensate for the loss of a vital bodily function (Watson *et al*, 2002). Home-based care for children with complex needs involved both a loss of privacy for families, and the loss of family space as the equipment and supplies necessary for the child's care consumes shared living environments. Privacy, safety, comfort and control remain valued features of home. Dyck *et al* (2005) suggest that the home is reconstructed physically, socially and symbolically and its primary meaning is contested when it becomes a site for regular long-term healthcare and associated services provided by outside paid workers. They suggest a blurring of the "private" and "public" space as the home is reconstructed as a space of caregiving and consumption. Similarly Wang and Bernard (2004) suggest that the intrusion of medical technology into the home has social and ethical consequences as it fundamentally alters the traditional meaning of "home". There has been no research conducted on this aspect of family life when a child has a disability and little empirical research on this aspect of caring for a child with a life-limiting condition or technology-dependence beyond a cursory mention in qualitative studies (O'Brien, 2001; Kirk & Glendinning, 2004; Steele & Davies, 2006; Carnevale *et al*, 2006, Ouelette, 2009).

In the context of children who are dependent on technology, studies report that that the home environment is organised to accommodate the needs of the child and the child's caregivers with parents describing how the equipment necessary for the child's care effectively took over the physical space of the home and medicalised the home environment (O'Brien, 2001; Kirk & Glendinning, 2004). Both of these studies suggest a significant loss of privacy for the family due to the significant movement of in-home support staff through the child's home. Loss of privacy was also an issue of concern for families in Steele and Davies (2006) study of children with progressive life-limiting conditions, in which parents described always having to be appropriate, in manner and dress, in their own home. Parents of children dependent on technology in a study by Kirk, Glendinning and Callery (2005) reported that technology transformed the meaning of home, which becomes medicalised by the presence of

equipment and the continual or frequent presence of home carers or professionals. Carnevale *et al* (2006) also report on the loss of the home environment to the medical equipment required for the child's care, or in some instances the adaptation of the house to accommodate this equipment. This qualitative study of the families of technology dependent children described the considerable lengths that parents went to in an attempt to camouflage medical equipment and subsequently make their homes appear more "normal".

Findings of qualitative studies exploring families of children with life-limiting conditions or technology-dependence are consistent with regards to the impact of medical technology on the home environment. Although these studies were not specifically focused on the population of interest in this study, and none were carried out in an Irish context, the findings are likely to be generalisable to this population in that the technology is essentially separate to the child and family and it was the impact of this, rather than aspect of the child or family, that has been described.

3.5.5 Financial Impact on the Family

Studies specifically exploring the financial impact on families of childhood life-limiting conditions are scarce, although Dobson *et al* (2001) suggests that it costs approximately three times as much to raise a child with any severe impairment as it does to raise a healthy child. Comparative studies suggest increased costs associated with caring for children with life-limiting condition (Monterosso *et al*, 2007) and children dependent on technology (Thyen *et al*, 1999). In a comparative study of parents caring for children with cancer (n=19) and parents caring for children with non-malignant life-limiting conditions (n=110) in Australia, Monterosso *et al* (2007) reported only a small difference in the rates of subjectively experienced financial strain between the two groups which was not statistically significant. This finding is possibly to be expected since all children were recruited to the study on the basis that they had a life-threatening condition and it may have been more meaningful to compare the group with healthy children. The authors also report that although both groups spoke frequently about difficulties in procuring funding for various forms of care, parents from the non-cancer group especially spoke of the burden they endured as a consequence of the lack of financial and practical assistance. Similarly in the USA, Thyen *et al* (1999) reported that although mothers of children dependent on technology reported higher out-of-

pocket expenses, the difference in the financial impact of caring for a child was insignificant between 65 mothers of children assisted by technology and 54 mothers of children hospitalised for acute illness. Although the study by Thyen *et al* (1999) focused on children who were technology-dependent, the findings are unlikely to be transferable to the population in this study in the context that a wide age range of children was included in the sample, and the study was conducted in the USA where systems and funding of care are significantly different to the Irish context. Similarly, despite the fact that over half of the children who had a non-malignant life-limiting condition in the study by Monterosso *et al* (2007) were categorised as having severe neurological disability, neither the nature of this, nor the age range of the children in the sample is specified. Consequently the generalisability of the findings to young children with life-limiting neurodevelopmental disabilities is not clear.

Conversely, in the context of caring for a developmentally disabled child in the U.K, Emerson (2003) conducted a comparative study of families of children with intellectual disability and families who did not have a developmentally disabled child. This study reported that on all indicators of socio-economic position (social class, jarman quintile, education and income) families caring for a child with intellectual disability were less advantaged than families who did not have an intellectually disabled child. One of the major limitation of Emerson's (2003) study was the lack of a formal definition of intellectual disability with the study identifying children as intellectually disabled based upon mothers' descriptions of their child as having "learning difficulties". This would suggest a very broad range of levels of disabilities amongst the sample. Additionally, the exclusion of children under the age of five, and the inclusion of children an associated ICD-10 psychiatric disorder, may have severely limited the generalisability of the findings to population of interest in the current study. Other researchers have explored the relationship between having a child with intellectual disability and the employment patterns of parents. Olsson and Hwang (2006) conducted a comparative study of wellbeing, involvement in paid work and division of child-care labour in Sweden. This study compared 179 families in which a child had intellectual disability with 196 families in a control group matched for age (<5 years). The study reported that both mothers and fathers in families where there was an intellectually disabled child worker fewer hours in paid employment than mothers and fathers in the control group families. In addition in the families where there was a child with intellectual disability the

mother worked fewer hours than the father. Although this study suggests a negative financial impact on families in which a young child has an intellectual disability due to reduced employment in paid work, it is difficult to generalise the findings beyond the context in which the study was undertaken. Sweden has a particularly well developed social welfare system and is unique with regards to support provided for parents to combine work and family.

Studies of families of children with complex health care needs also report increased financial burden for families. In a review of research literature published between 1989-2005 Anderson *et al* (2007) conclude that the financial burden incurred by the families of these children can be substantial, especially among families who care for a child with a severe disability. Other studies have investigated the relationship between healthcare expenditure and perceived financial burden for families (Chen & Newacheck, 2006), child health status (Kuhlthua *et al*, 2005) and child and family characteristics (Lindley & Mark, 2010). While these studies consistently report increased financial burden for families, particularly in association with increasingly complex need and reduction in parental employment hours, the fact that they were conducted in the USA limits the generalisability of the findings beyond that particular context.

Similarly, findings from a range of descriptive studies also tend to be consistent in terms of describing a negative financial impact for families. Lenton *et al*'s (2001) cross-sectional survey of 93 families of children with a wide variety of life-limiting illnesses, reported that 81% of the families reported financial difficulties, although the exact nature of these difficulties are unspecified. All families of children who were dependent on technology in O'Brien's study (2001) described additional financial costs associated with their child's condition. For families in this study these tended to be intangible costs associated with the extra money needed to run the household (for example electricity and heating). Similar findings were reported by Quinn *et al* (2005) in their study of Irish families of children with a wide range of life-limiting conditions requiring palliative care. Families in this study also reported increased costs associated with one parent having to give up work to care for the child, and costs associated with having to spend long periods away from home when the child is hospitalised. The mothers in Redmond and Richardson's Irish (2003) study also reported

increased financial costs associated with having to buy essential services and equipment. Negative financial impact was also reported by the ten families in Steele and Davies's (2006) study of families of children with progressive life-limiting illnesses. Again this study suggested this was principally associated with giving-up or reducing outside employment, although families also reported additional "hidden costs" associated with extra heating, extra laundry and special equipment needed for the child.

Although contributing to our knowledge of the experience of caring for children with complex needs, quantitative assessments of the financial burden experienced by these families are limited in their generalisability beyond the particular context in which they were undertaken. Alternatively while research studies demonstrate an association between families of children with intellectual disabilities and low income the design of these studies means that the association might work either way. However, findings from qualitative studies suggest that families of children with life-limiting conditions and disabilities do suffer a negative financial impact as a consequence of the increased intangible costs of providing ongoing care. These include loss of employment opportunities and the additional costs associated with maintaining the household and purchasing items and equipment necessary for the child's ongoing care.

3.5.6 Impact on Parents of Providing Care

Parents who live with and care for a child with severe and complex health conditions face a considerable number of different stressors. Researchers have concluded that parents whose children have any type of disability or illness report higher levels of emotional and physical stress than parents whose children are healthy (Carnevale *et al*, 2006, Dellve *et al*, 2006). Hauskov-Graungaard *et al* (2011) suggest that the experience of having a child with a severe and complex disability is not a single stressor, but an ongoing situation with continual new challenges and difficulties. The stresses that families caring for children with life-limiting illnesses experience are neither fixed nor predictable (Ouellette, 2009). They coalesce in patterns that seem to be family specific because different families view the same stressor very differently (Ratliffe *et al*, 2002). Much of the research related to the impact of caring for a child with a life-limiting condition or developmental disability focuses on the psychological impact on the mother, possible because mothers are more likely to be directly involved in the

child's routine medical care (McKeever & Miller, 2004; Redmond & Richardson, 2003; Green, 2007a; Monterosso *et al*, 2007; Manaseri, 2008).

3.5.6.1 Psychological Impact of Care Provision

Studies measuring the psychological impact of providing care specifically to children with life limiting conditions are relatively rare, with only three studies located, one of which was conducted in an Irish context.

Lenton *et al* (2001) reported a prevalence rate of psychological morbidity of 54% for mothers and 30% for fathers of children with life-limiting conditions in their cross-sectional survey in the U.K. Psychological distress amongst the parents sample was measured using the General Health Questionnaire [GHQ]. However, neither the version of the GHQ nor the threshold used to identify psychological morbidity were specified. This study included a wide range of children with both malignant and non-malignant diagnoses although no distinction was made between the groups. Later studies suggest differences in prevalence of psychological morbidity between parents of children with cancer and non-malignant life-limiting conditions. In an exploration of the supportive and palliative care needs of children with life-threatening illness in Australia, Monterosso *et al* (2007) used the 14-item Hospital Anxiety and Depression Scale to compare rates of psychological distress between mothers caring for a child with cancer and mothers caring for a child with non-malignant life-limiting conditions. This study reported a high incidence of moderate to severe anxiety (79%) and depression (68%) amongst the mothers of children with cancer compared to a rate of 34% of moderate to severe anxiety and 23% of depression amongst the mothers of children with non-malignant life-limiting conditions. Although both of these studies suggest higher levels of psychological distress amongst parents of children with life-limiting conditions it is impossible to compare the findings beyond this in any meaningful way. The studies were conducted in two different social and service contexts, and used two different measurement instruments. Their relevance to the current study lies in the inclusion of children with complex life-limiting disability amongst both samples. However Monterosso *et al* (2007) provide no details on the age range of children in their sample, and Lenton *et al* (2001) included a wide spectrum of ages in theirs.

Alternatively, Redmond and Richardson (2003) used the 12-item GHQ to assess psychological morbidity in a sample of 17 mothers of severely / profoundly and life-limited intellectually disabled children, under four years of age in Ireland. The authors report that 88% of the mothers were experiencing elevated levels of strain, with 71% reporting recent higher levels of unhappiness and depression. This study is limited not only by the small purposefully selected sample, but also by the fact that the authors base these estimates on single item responses to the GHQ rather than the scales recommended threshold for identifying overall psychological distress. Despite this, it is of particular relevance to the population of interest in the current study in the context that it is the only study to focus specifically on the same population and age range of children, and was conducted in the same cultural and service context as the current study.

In contrast to children with life-limiting conditions, studies exploring the psychological impact of caring for a child with intellectual disability are plentiful. The findings of these studies are consistent and report elevated levels of psychological morbidity amongst parents of children with intellectual disability compared to a norm reference group of parents (Seltzer *et al*, 2001; Emerson *et al*, 2006; Olsson & Hwang, 2006; Singer *et al*, 2006; Bailey *et al*, 2007; Thurston *et al*, 2011). However studies that have specifically explored gender differences in the experience of stress between mothers and fathers of children with intellectual disabilities suggest that the experiences of fathers and mothers differ, with mothers more likely to feel intense stress and experience emotional distress than fathers (Olsson & Hwang, 2002; do Amarail, 2003).

Emerson (2003) used the GHQ12 in association with a self-report psychological impact scale to measure psychological distress in a comparative study of 245 mothers with an intellectually disabled child, aged 5 – 15 years, and a group of mothers of non-disabled children. This study reported a prevalence rate for psychological morbidity of 35% amongst the mothers of children with intellectual disability compared with a rate of 25% of mothers in the comparison group. The study also identified a number of associations between the assessed mental health status of the sampled child's mother including indicators of deprivation, the number of potentially stressful life events experienced by the child, the mother's self-assessed social impact of the sampled child's difficulties, family functioning,

and the gender of the sampled child. The degree to which this study can inform the current one is difficult to estimate. Emerson's (2003) study excluded children in the age range to be investigated in the current study, and relied upon the use of an un-validated operational definition of intellectual disability which encompassed children with a wide range of levels of intellectual disability.

Olsson and Hwang's (2006) comparative study of 179 families in which a child had an intellectual disability and 196 control families used the Beck Depression Inventory [BDI] to examine psychological wellbeing between mothers and fathers of children with an intellectual disability, and between parents of children with an intellectual disability and a norm reference group. In this study mothers of children with intellectual disability reported more depressive symptoms than fathers, and parents of children with intellectual disability reported more depressive symptoms than parents in the control group. Although this study reported a positive relationship between involvement in paid work and wellbeing for both mothers and fathers of children with intellectual disability, the cross-sectional nature of the survey means it is impossible to distinguish whether poor psychological wellbeing was the reason for low employment, or whether employment had a positive effect on psychological wellbeing in the study group. Although the findings were consistent with those of Emerson (2003), again the degree to which this study can inform the current study is difficult to estimate. Like Emerson (2003), Olsson and Hwang (2006) included children from birth to 16 years, with a wide range of intellectual disabilities ranging from mild to severe.

Similar findings are reported by Seltzer *et al* (2001) who conducted a comparative study of three groups of parents of children with a developmental disability, a serious mental health problem, and a normative group to explore life-course impacts of caring for a child with a disability. This study confirms other reports that parents of children with developmental disabilities worked fewer hours in paid employment and had reduced rates of social participation over the life course. However parents in this group did not differ from the comparison group in terms of physical wellbeing, which was assessed using a self-rating scale, or psychological wellbeing which was assessed using the Center for Epidemiological Studies Depression Scale. The unique feature of Seltzer *et al*'s study (2001) was that the participants were selected for the larger study prior to and independent of the birth and

diagnosis of their child. Although this study was unique in its ability to circumvent self-selection bias and provide information on pre-existing variables in the sample, the degree to which it can inform the current study is limited by the fact that it focuses on the parents of adults with intellectual disability.

More recently Thurston *et al* (2011), in a cross sectional survey of parents and guardians of children with complex special needs, reported that 41.6% of the carers in their study exhibited symptoms (mild to severe) of psychiatric distress using the Kessler scale as a measurement tool. The presence of psychological distress was associated with reports of poorer social support, family dysfunction, greater adverse impact of the child's situation on the family, poorer child behaviour, unfavourable parenting styles and poorer child psychosocial functioning. The severity of the child's physical dysfunction was not related to parents' / guardians symptoms of psychological distress. The extent to which this study can inform the current one is limited by the fact that the nature of the child's complex needs is not defined, and families of children from 16 different ICD-10 diagnostic categories were included in the sample. In addition although 40% of the children in the sample were aged less than five years, the sample included children aged up to 19 years.

Two studies have used meta-analysis to explore literature related to the psychological impact on parents of caring for a child with a developmental disability with reasonable consistent findings. Singer *et al* (2006) conducted a meta-analysis of 18 studies of depression in mothers of children with and without developmental disabilities. This meta-analysis included only studies in which data was collected using standardised self-report measures with well established psychometric properties. The analysis found higher levels of depressive symptoms in mothers of children with developmental disabilities, with an effect size of .39 and estimated average of 29% of mothers with significantly elevated symptoms. Similarly Bailey *et al* (2007) conducted a meta-analysis of 42 studies specifically measuring depressive symptom in parents of children with intellectual disability. This analysis included studies in which depression was diagnosed using a psychiatric interview applying DSM-IV criteria. Computing a weighted average of reported rates of individuals who passed a screening threshold for current depression, Bailey *et al* (2007) reported an estimated rate of 23.6% of

significantly elevated symptoms of depression, slightly lower than the figure reported by Singer *et al* (2006).

In summary, numerous studies have reported elevated levels of psychological distress amongst mothers, and to a lesser extent fathers, of children with life-limiting conditions and intellectual disabilities. Although the findings of these studies are generally consistent, there are several difficulties associated with comparing and contrasting this research literature, which principally relate to the constructs and methodologies used. Studies have assessed different samples of parents, of different populations of children, at different ages and developmental stages. In addition samples are often not rigorously constructed, and the wide variety of measurement scales and instruments used makes comparisons of prevalence rates between studies difficult, and may account to the differences reported. Despite these difficulties studies generally report higher rates of distress and lower rates of well-being among mothers, and occasionally fathers, of children with life-limiting conditions or developmental disabilities. The difference in levels of psychological distress between mothers and fathers may perhaps be explained by the more demanding parental role in which mothers are involved, and the fact that as mothers consistently worked fewer hours in paid employment they may have been more socially isolated than fathers. The degree to which these studies can inform the conduct of the current study is limited by several factors including the broad age range of children in the study samples which makes application of the findings to a particular age group of children difficult. Additionally, the fact that children with life-limiting disabilities are subsumed into the samples of studies exploring life-limiting conditions more generally means that the relevance of these general findings to this specific population of children and their families is difficult to estimate. Finally, studies specifically exploring children with developmental disabilities have either used un-specified classifications of intellectual disability, or have included a broad spectrum of level of intellectual disability, consequently the applicability of the findings of these studies specifically to children with life-limiting disabilities is uncertain.

3.5.6.2 Physical Impact of Care Provision

The physical impact of caring for a child with a life-limiting condition or developmental disability has also received attention in the research literature in recognition of the extra

physical burden on carers. Three comparative studies could be found which explored this aspect of care, although all focused primarily on the mother. Thyen *et al*'s (1998) study of 65 mothers of children assisted by technology and 54 mothers of children hospitalised for acute illness reported impaired general health related to pain, vitality, and social functioning in mothers of the technology-dependent children. At a more general level, in their comparative study of mothers caring for a child with cancer and mothers caring for a child with non-malignant life-limiting conditions Monterosso *et al* (2007) reported that 95% of mothers caring for children with cancer described their general health as good or excellent general health compared with 61% of the mothers caring for children with non-malignant life-limiting conditions. They suggest significant differences in the carer burden for the two groups including a much longer caring trajectory, more severe disability and a higher carer burden amongst the non-cancer group of children account for the difference between groups. Similar findings are reported by Brehaut *et al* (2009) in a large scale population-based Canadian study which compared the health of 3633 caregivers of healthy children with the health of 2495 caregivers of children with a variety of chronic and disabling conditions. The children with chronic conditions ranged in age from 4-11 yrs. and 90% of the primary caregivers were the child's mother. Consistent with the findings from previous studies Brehaut *et al* (2009) report significantly poorer health amongst the caregivers of children with chronic health problems after controlling for family, caregiver and child related factors.

The principle difficulty associated with comparing the findings of quantitative studies on parental health relates to the instruments used to measure the construct. Thyen *et al* (1998) used a subscale of the Center for Epidemiologic Studies Depression Scale, however neither Monterosso *et al* (2007) nor Brehaut *et al* (2009) specify a validated measurement instrument. Although these studies report consistent findings the transferability of these to the population of interest in the current study is not certain. Neither Thyen *et al* (1998) nor Monterosso *et al* (2007) specify the age range of children in their studies, while Brehaut *et al* (2009) excluded children in the age range that this study is concerned with.

The physical impact of providing care to children with life-limiting conditions has also been briefly described in qualitative studies. These consistently describe two main physical impacts, namely parental fatigue and exhaustion (O'Brien 2001; O'Brien & Wegner, 2002;

Kirk & Glendinning, 2004; Greene 2007a; Monterosso *et al*, 2007; MacDonald & Callery, 2007; Smith Stepanek, 2008). Steele and Davies' (2006) study reported that fatigue was a particularly prevalent symptom that held the potential to impact on parents in multiple ways. Parents in this study of children with life-limiting neurodegenerative conditions also reported a range of somatic symptoms including injuries, migraine headaches, and anaemia. Similarly, Smith Stepanek (2008) reported that cognitive and physical exhaustion was a part of daily life for parents caring for children with life-limiting conditions. While specifically in a disability context, Murphy *et al*'s (2006) qualitative exploration of the physical and emotions health of caregivers of children with intellectual disabilities participants reported that they spent on average 14.7h / day in caregiving activities, and describe physical problems that included chronic fatigue, sleep deprivation, pain and injuries. The inclusion of both young and adult children in this study limits the extent to which the findings can be generalised specifically to young children with life-limiting neurodevelopmental disabilities.

3.5.7 Impact on Siblings

Living with a sibling with a life-limiting condition clearly also has consequences for other children in the family, and well siblings of children with these conditions have unique needs of their own which also require attention, understanding and support. Besier *et al* (2010) suggests that healthy siblings of chronically ill children face multiple challenges such as exposure to the physical and emotional pain of their sibling's condition, fear, parental distress, and extended separation from their sibling and parents because of hospitalisations. Using standardised assessment tools such as the Sibling Perception Questionnaire (Sloper & While, 1996; Stallard *et al*, 1997; Lenton *et al*, 2001), and the Strengths & Difficulties Questionnaire (Taylor *et al*, 2001; Besier *et al*, 2010), studies report a prevalence rate of 20 – 30% of poor adjustment in siblings of children with life-threatening conditions. Moreover, parents often consider that their concerns about their healthy children's adjustment are an additional stress factor in family life (Eiser, 1993; Manaseri, 2008).

Studies on well siblings of children with life-threatening or life-limiting conditions have tended to be quantitative in nature and focused on the measurement of well siblings' adjustment. Studies that have used the Sibling Perception Questionnaire (Stallard *et al*, 1997; Lenton *et al*, 2001) have reported high rates of sadness and emotional or behavioural

problems, and the considerable communication needs of healthy siblings of children with life-limiting conditions. There is also evidence that parents overestimate to degree to which healthy siblings cope (Lenton *et al*, 2001). Taylor *et al* (2001) provided additional support for these findings using the Strengths & Difficulties Questionnaire to explore the adjustment of siblings of children with a wider range of malignant and non-malignant illnesses requiring daily routines of care. This study reported no significant association between ill siblings' malignant or non-malignant diagnosis and their healthy siblings' total difficulty scores with emotional subscale scores elevated for all healthy children, and significantly associated with maternal emotional distress. More recently Besier *et al* (2010) expanded upon the findings of Taylor *et al* (2001) by using the SDQ to compare siblings of children with a chronic, life-limiting or life-threatening illness with a matched control group of children from the general population. This study also reported an elevated risk of having emotional and behavioural problems in the study group, with no significant difference in any of the subscales or total scores between the siblings relative to the diagnostic groups of the ill children.

The consistency of the findings of quantitative studies exploring well sibling of children with chronic, life-limiting and life-threatening conditions suggest a negative impact on well siblings emotional health. The findings also suggest that this impact is similar regardless of the nature of the ill child's condition or diagnosis. However, at a general level these studies suffer from a number of methodological weaknesses. Studies have relied upon volunteer families and it is unclear whether such samples are representative of the general population or biased towards better adjusted families who may be more willing to participate. In addition most have relied upon parents reports of sibling adjustment and perceptions (predominantly mothers), which have been demonstrated by Lenton *et al* (2001) to be inconsistent with the self-reports of well siblings. Typically studies have included children of all ages making it difficult to account for developmental stages in well children's adjustment.

In contrast to studies exploring the well siblings of children with life-threatening or life-limiting conditions, studies exploring the siblings of children with intellectual disabilities have tended to be predominantly qualitative in nature and focused on the perceptions of well sibling. In a grounded theory study exploring children's perceptions of their intellectually disabled siblings Stalker and Connors (2004) conclude that while children were aware of

their sibling's impairments with varying levels of understanding, the majority did not construe this as marking their sibling as "different". Stalker and Connors (2004) conclude that children's perceptions were dominated by other people's reactions. Similar findings were reported in a study by Warren-Dodd (2004) which also highlighted varying levels of understanding of their sibling's condition, although unlike the children in Walker and Connors' study (2004), well siblings in Warren-Dodd's study (2004) reported both positive and negative perceptions of their disabled sibling as did the well siblings in a study by Naylor and Prescott (2004). It is worth noting that in both of these studies the primary focus was on the evaluation of a support group for well siblings. The degree to which this may have influenced the findings is not clear.

The findings of qualitative studies exploring the perceptions of well siblings of children with disability tend to be consistent, and suggest that well siblings of children with intellectual disabilities report both positive and negative perceptions of their siblings. However these perceptions tend to be focused on the sibling and not on the disability or illness per se, and in general these were probably not that different from the views that any child would express when asked about their siblings. Like the quantitative studies exploring sibling adjustment studies are limited by small numbers and convenience samples which included children of all ages, again making it difficult to account for developmental changes in children's perceptions. In a review of sibling disability literature Stoneman (2005) concludes that there is a common theme across the issues in need of additional research, namely, the importance of developing large sample and multisite studies of siblings as the issues are complex and cannot be adequately addressed when sample sizes are small.

There may however be more subtle impacts on well siblings of children with life-limiting conditions and disabilities that are not accounted for by children's perceptions and levels of adjustment. Parents' emotional and physical availability to their healthy children is important (Bradford, 1997; Taylor *et al*, 2001) and several studies have commented on this aspect of life for a well sibling of children with technology-dependence and children with complex disability. While some children in Naylor and Prescott's study (2004) reported that they were treated the same as their sibling with a disability others reported a lack of attention from their parents because their sibling needed more help and support from carers. This may account for

the findings of Sharp and Rossiter's (2002) meta-analysis of sibling impact that illnesses with daily treatment regimes were associated with greater negative effect than those that did not affect daily functioning. Similar findings were reported in the context of technology-dependent children by O'Brien (2001) and Carnevale *et al* (2006) and for children with life-limiting conditions (Manaseri, 2008). Parents in O'Brien's study (2001) also describe how the nature of the care required by their technology-dependent child leaves little time to devote to the needs of other family members. Parents in Carnevale *et al* (2006) also described how siblings are neglected as a consequence of the time that needed to be devoted to the technology-dependent child. Many parents report this as an additional source of stress, anxiety, and grief. Carnevale *et al* 2006; Manaseri, 2008; Ouelette, 2009)

It would also appear that social opportunities and interactions for well siblings also suffer. Nicholl (2008) reports limited social opportunities for siblings of children with complex needs as a result of the attendant practical difficulties associated with leaving the home environment, while Carnevale *et al* (2006) report similar difficulties in the context of children dependent on medical technology. In the context of childhood disability the issue appears to be more complex and less concerned with the pragmatic aspects of caregiving. Parents in Burke's (2010) mixed methods triangulation study reported limited opportunities for doing things together as a family, while well siblings in Stalker and Connors' study (2004) also report the loss of normal social opportunities as parents insist on including their disabled child in all social occasions. However while Stalker and Connors (2004) report that disability is constructed as a form of normality within the family itself, Burke (2010:1694) proposes that the experience of well siblings is essentially "disability by association" in their social encounters.

In summary, despite consistent findings across studies the issue of psychological impact on well siblings of children with a chronic, life-limiting conditions, or intellectual disabilities appears to be complex. In the face of findings from quantitative studies 70-80% of well siblings are well adjusted, and do not report significant difficulties. This is reinforced by meta-analyses of sibling impact literature. In a meta-analysis of 25 studies on the siblings of individuals with intellectual disabilities Rossiter and Sharpe (2001) reported only a small negative effect for having an intellectually disabled sibling. This negative effect was more

pronounced for measures of psychological functioning, and adult reports versus child self-report. On repeating the meta-analysis procedure with siblings of children with a variety of chronic illnesses Sharpe and Rossiter (2002) report a modest negative effect relative to comparison participants or normative data. Again this negative effect was more pronounced for measures of psychological functioning, and adult reports versus child self-report. However illnesses with daily treatment regimes were associated with greater negative effect than those that did not affect daily functioning.

3.5.8 Summary of Research Literature Related to the Demands and Risk Factors for Family Adjustment

There is a considerable body of knowledge related to the demands placed on families caring for a child with a life-limiting condition or developmental disability. The research literature suggests that family caregiving in this context is associated with a variety of variables that contribute to the overall stress experienced by families. These include factors related to the nature and management of the child's care, balancing family life, and a variety of negative impacts on both individual family members and the family as a unit. According to Family Stress, Adaptation and Adjustment Theory (McCubbin & Patterson 1983a, 1983b; McCubbin & McCubbin, 1993) these factors do not function in isolation but rather act synergistically to influence family outcomes.

However family outcomes are not determined solely by the accumulation of these considerable demands. They are also influenced by a variety of mediating and resource factors which help ameliorate the stress and burden experienced by families. The following section of this research literature review focuses on these intra-familial resources, and these services which are available to families to support them in their caring role.

3.6 Family Resources and Intervening Factors: bB Factors

It is a major tenet of Family Stress, Adaptation and Adjustment Theory that all families have resources and capabilities for meeting the demands of a crisis situation, and that these include characteristics of individual family members, the family unit, and the community of which

the family is a part (McCubbin & Patterson 1983a, 1983b; McCubbin & McCubbin, 1993). The theory proposes that when families have the resources they need they are better able to adapt to adversity by coping with the imbalance between the demands of the stressor and their ability to respond and minimise the attendant distress. The model identifies both intra-familial and external resources which influence family coping in childhood illness.

However, in the context that this study was not designed to be a test of the model intra-familial resources are not explored in this study and consequently are not reviewed in depth in this section of the research literature review. There is a vast volume of literature related to family stress and coping in the context of childhood intellectual disability which has used various theoretical frameworks to examine these phenomena (although this is not so prevalent in the context of childhood life-limiting illness). Generally studies have focused on identifying the relationships between variables associated with certain child characteristic, family characteristics and the availability of formal support systems with family stress and coping; or on differences between mothers and fathers perceptions of stress and coping strategies. (Heaman, 1995; Hoare *et al*, 1998, Smith, Oliver & Innocenti, 2001; Olsson & Hwang, 2002; Lam, Giles & Lavander, 2003; Kelso, French & Fernandez, 2005; Glidden, Billings & Jobe, 2006; Giallo & Gavidia-Payne, 2006; Glidden & Natcher, 2009; Larson, 2010). Studies suggest that, in families of children with disabilities, parental stress shows a stronger statistical relationship to family cohesion, income level, and family and social support than to aspects of child functioning and severity of disability. Although extensive, this literature is likely of limited relevance to the specific population that this study addresses. These studies have approached intellectual disability as an umbrella term, and subsequently included children of all functional levels including those with associated psychiatric disorders and challenging behaviours. In addition many studies have used the term “child” to refer to a relationship rather than an age category and consequently include both young and adult children further limiting the extent to which they can inform the current study.

In the context of childhood chronic, life-limiting or severely disabling conditions, studies have identified a variety of factors associated with parental stress and coping resources. These include: access to information (Lenton *et al*, 2001; Kirk & Glendingning, 2002;

Redmond & Richardson, 2003; Quinn *et al*, 2005, Monterosso *et al*, 2007; Smith Stepanek, 2008), the degree of uncertainty in the child's chronic condition (Dodgson *et al*, 2000; Cohen, 1993), family cooperation and social and emotional support (Taanila *et al*, 2002; Katz, 2002), family function (King *et al*, 1999), continuity of care and relationships (Heller & Solomon, 2005) and feelings of personal competence and control (Dellve *et al*, 2006; Llyod & Hastings, 2009). There is also evidence that mothers and fathers experience family stress to different degrees (Katz, 2002; Dellve *et al*, 2006), report different experiences of burden, meaning and growth (Ware & Ravel, 2007; Kennedy, 2009; Schneider *et al*, 2011), and favour the use of different coping resources and strategies (King *et al*, 1999; Swallow *et al*, 2011).

This section of the research literature review focuses instead on services to children and their families since this was the main intervening factor addresses by this study. Formal service support is identified in Family Stress, Adaptation and Adjustment Theory as a critical family resource in the context of childhood illness (McCubbin & Patterson 1983a, 1983b).

Subsequent research in families of children with life-limiting conditions and families of children with severe disabilities support this position reporting that support from formal service systems is a significant and important parental resource for family coping and adaptation (King *et al*, 1999; Katz, 2002; Taanila *et al*, 2002; Kirk & Glendinning, 2004; Dellve *et al*, 2006; Kenny & McGilloway, 2007; Ouelette, 2009).

3.6.1 Parents' Experiences of Services to Children and their Families

Access to appropriate health-care services is vital for the health and wellbeing of all children. While children with complex neurodevelopmental disabilities have the same range of need for services and support as other disabled children, their medical, physical social and emotional care requires proportionally more time, physical and financial resources (Nolan *et al*, 2005; Campbell, 2007), and they are likely to be in contact with many different agencies and services (Cass *et al*, 1999; Redmond & Richardson, 2003; Quinn *et al*, 2005).

Literature related to service access for children with special or complex medical needs, of which children with life-limiting conditions form a specific subgroup, is almost entirely

dominated by American studies (Newacheck *et al*, 2000; Wyngaarden Krauss *et al*, 2003; Mayer *et al*, 2004; Erickson-Warfield & Gulley, 2006; Bitsko *et al*, 2009; Hefner, 2010). The findings of these studies are consistent, and suggest access difficulties and considerable unmet service needs amongst this population of children. They also report increasing access difficulty as the child's needs become more complex or their condition more unstable, or where the parent was in poor health. However, the context in which these studies were undertaken means that the findings are unlikely to be generalisable to the Irish context with its free access to publically funded health services. In addition these studies have tended to focus on service access as a binary outcome; this does not take account of the many difficulties that parents experience in trying to access the services in the first place.

Alternative qualitative studies have tended to focus simultaneously on issues of access to services and the process of service delivery. As part of an exploration of the palliative care needs of children with a wide variety of life-limiting conditions in Ireland, Quinn *et al* (2005) explored parents' experiences of service availability and service usage. The 34 purposefully selected parents in this study reported insufficient community based services, and considerable difficulty in accessing essential services and equipment. They also reported unnecessary bureaucracy, lack of information and delays in service provision. Experiences of hospital based services were generally positive, although some parents were critical of the poor communication skills of hospital personnel and the physical conditions encountered in hospital settings. Parents in this study emphasised the value and importance of home care and respite services, and the significant role of the voluntary sector in the provision of services. Although this study had some relevance to the population of the current study in that it was conducted in an Irish context, and used a nationally representative sample, the broad range of diagnostic categories and range of ages of the children in the study (0-27) make it impossible to distinguish the service needs of any specific groups of children and their families. However the findings support previous research by Redmond and Richardson (2003), also conducted in Ireland, but this time focused specifically on young children with severe and life-limiting disabilities. Mothers in Redmond and Richardson's study (2003) also reported services that were insufficient, inconsistent and inadequate to meet the child's needs. Similar to Quinn *et al* (2005) difficulties with bureaucracy and delays in obtaining necessary services were common, with mothers describing uncoordinated, disconnected services that were an

additional source of tension and strain. The consistence of these findings would suggest that these difficulties are uniformly experienced by all families regardless of the child's age or diagnostic category.

Studies from the U.K. are also consistent, and report findings similar to those conducted in Ireland. Kirk and Glendinning (2004), in their qualitative study of services and supports for parents caring for a technology-dependent child at home, describe community based services that were not sufficiently developed to support these families. Generally services were poorly planned and coordinated which resulted in families receiving services from a number of organisations and professionals who were sometimes confused about the responsibilities and roles of different support services. While in a mixed methods study Hunt, Elston & Galloway (2003) explored perceptions of services for children with palliative care needs from the perspective of a mixed group of service users including parents, carers and young persons (n=272) and also reported services that were difficult to access and which lacked resources and funding. Consistent with previous descriptive reports this study found a large proportion of parents (47%) reported services that were needed but were unavailable or difficult to access, with 58% of parents reporting that they met barriers when trying to access care and support. Principle amongst these was a lack of resources and funding. Consistent with Irish studies (Redmond & Richardson, 2003; Quinn *et al*, 2005) parents also described services that were characterised by delays, lack of information, and bureaucracy.

Despite being conducted in two different service contexts, studies from the U.K. and Ireland report consistent findings regarding service provision to children with life-limiting conditions or technology-dependency. These studies suggest parents experience a number of access and process difficulties with services that are characterised by inconsistency and underfunding. The one study that specifically focused on young children with life-limiting neurodevelopmental disabilities suggests that these are also experienced by this group of children and their families. These studies provide a rich description of perceptions and experiences of services available to children and families, although they lack any particular focus for intervening to improve these services.

Other qualitative studies, although focused more generally on the overall experience of caring for a child with a life-limiting condition or complex medical needs, also provide descriptions of parents' experiences of engaging with the services available to their child and family. For the most part these qualitative studies describe process rather than access variables, and once again findings are consistent, regardless of the context in which the study was undertaken. Carnevale et al (2006) describe the impact of limited resource allocation on both the child who is technology-dependent and the family, and report services which are difficult to obtain and characterised by long delays. Similarly Nicholl (2008) reports that getting services was difficult, and keeping them problematic, for the mothers of children with complex needs in her study. Mothers of children with life-limiting illnesses in Ouelette's (2009) describe several process difficulties with healthcare providers including communication difficulties and information difficulties. Manaseri (2009:143) describes the services to mothers of children with complex medical needs in her study as "characterised by a rhetoric of choice and integration" as mother sin this study discuss the difference between "on-paper" services and "real-life" services. While these studies provide a rich description of perceptions and experiences of services available to children and families they are subject to the limitations of any small scale study with a purposefully selected sample. In addition many lack a particular focus for intervening to improve these services.

In summary, literature focusing on issues of services for children with life-limiting conditions and complex disabilities from the perspective of parents report consistent findings, regardless of the service context in which they were undertaken. Studies suggest that parents are consistently confronted by issues of access, insufficient services, poor coordination and integration, beurecracy and delay.

3.6.2 Service Providers Perceptions of Services to Children and their Families

Hunt, Elston & Galloway (2003) and Quinn *et al* (2005) also explored service providers' perspectives on the services available to children with life-limiting conditions as part of their studies in the U.K. and Ireland respectively.

In a study described as a mixed methods study but essentially a postal survey with open ended questions, Hunt, Elston & Galloway (2003) explored perceptions of services for children with palliative care needs from the perspective 144 health care providers (health professionals / social care workers / support workers). Consistent with service users, service providers (58%) also reported services that were needed but were unavailable or difficult to access. One of the main strengths of this study was its comparison of the views of parents and service providers, which demonstrated both agreement and disagreement on individual aspects of services. The identification of some aspects of unmet needs were consistent between the groups with both groups identifying insufficient respite services and insufficient emotional and social support for parents. However there were also come conflicting opinions on some aspects of services available: for example, although service providers considered that only 2% of parents did not have an identified key-person to call upon to organise care or support, 46% of parents reported that this was the case. An additional strength of the study was its identification of solutions to the difficulties parents experience rather than simply describing these difficulties. Parents proposed that they needed more services, which were better coordinated, and a key worker who could help them coordinate and access the services they needed. Alternatively health professionals proposed joint directorates and shared budgets for agencies involved in the provision of care. One of the major limitation of this study was the low response rate of 30% overall (no individual figures are given for the separate groups) although this still represented one of the largest samples used in exploring services to this group of children and their families. In addition, all participants were recruited through the ACT database, subsequently only those registered with the database were included in the study.

In an Irish context Quinn *et al* (2005) also explored professional and voluntary workers perceptions of palliative care, and their experiences of providing palliative care for children with life-limiting conditions, although many of the participants did not acknowledge the care they provided as palliative care. Data was collected using postal questionnaires and during 15 focus group interviews with a broad range of professional disciplines and agency types, and from 12 individual interviews with key service providers and policy makers. The overwhelming view of service providers was for the need for a multidisciplinary team approach to service provision, that is flexible, and child and family centred. The need to

expand the services of the palliative home care team to include children with life-limiting conditions was considered a priority. Concern was expressed about the availability, consistency and equity of current services, in particular respite services and in-home support services were considered insufficient. Concern was also expressed about funding issues affecting the availability of essential equipment and appliances. The breadth and inclusive nature of this study may be considered both a strength and a limitation. It may be perceived as a strength in terms of its provision of a generic overview of the opinions and experiences of a wide variety of agencies involved in the care of a wide variety of children with a broad range of life-limiting illnesses. Alternatively it may be a potential limitation of the study in that many of the professionals and agencies involved have a specific and quite unique focus, differing political agendas, and a very wide range of unique experiences and expertise. The study reports the combined views of participants, however the demographic data reported in the study suggests considerable variations in the experiences of this disparate group of service providers in providing care to the population of children and families that the study explored.

Studies exploring service providers' perceptions of the services they provide to children with life-limiting conditions are relatively scarce. However, despite two different service contexts, the findings of these studies are remarkably consistent, both with each other and with parents reports. Service providers in both studies reported services that lacked resources and funding, and both identified unmet service needs that included insufficient respite services and insufficient emotional and social support for parents. In both studies service providers also suggested that accessing services and supports was difficult not only for parents, but also for service providers themselves.

3.6.3 Summary of Research Literature Related to Intervening Factors

A considerable volume of research literature exists related to family stress, coping and coping resources in the context of childhood intellectual disability and childhood life-limiting illness which identifies a wide variety of inter-familial resources which influence family coping and adjustment. One of the critical extra-familial resources that has been identified in the research literature is the issue of formal support from the services available to the child and family.

Although these studies provide an international perspective, which incorporates a variety of service delivery models, the findings are generally consistent across studies. Findings suggest that parents are constantly confronted with under-funded, inconsistent and uncoordinated services, which are difficult to access, and insufficient to meet the needs of children and families. Findings from studies which have explored the perspectives of service providers support parents' experiences and report similar experiences amongst the service providers themselves.

3.7 Meaning and Appraisal in the Care of the Child: cC Factors

Within Family Stress, Adaptation and Adjustment Theory "cC" factors refer to the meaning the family gives to the total crisis situation, including the stressor and the associated strains. Families' ability to successfully re-define a crisis situation and give it meaning is a critical component of family adjustment and adaptation (McCubbin & Patterson, 1983a, 1983b). This section of the research literature review focuses on the meaning families attribute to their situation and parents positive perceptions of their child.

3.7.1 Parents Positive Perceptions of their Child

A feature specific to the childhood disability research literature is the concept of positive family perceptions of children with disabilities, with perceptions of burden and stress associated with the care and parenting of children with disability much criticised in recent literature. Emerson *et al* (2006) criticises researchers for focusing on the emotional burdens of having a child with intellectual disability and less on the burdens imposed by the inadequate societal support for the time-consuming and expensive task of caring for such a child. Similarly Green (2007a) cautions researcher to remember that parents of children with disabilities raise their children within the context of a powerful societal discourse that devalues disabilities and they are expected to feel emotionally burdened. Summers, Behr & Turnbull (1989) suggest that it should be acknowledged that a child with a disability encompasses a multitude of characteristics, some related to the disability and some not, which may lead to stress in the family, while other characteristics of the child may yield non-

stressful effects that may be either positive or negative. In this regard, a child with a disability may be no different to children without disabilities.

There has long been evidence to support the fact that children with intellectual disabilities make a positive contribution to family life, although initially, in many studies the finding was incidental to the major interest of the investigation. Studies report several positive aspects to parenting a child with a disability including a closer and stronger family; personal growth, more patience and compassion, unselfishness; and a greater appreciation for the small and simple things of life (Wikler *et al*, 1983; Abbot & Meredith, 1986; Mullins, 1987). Hornby (1992) found that the majority of parents felt their lives were enriched and made more meaningful, regardless of the type or severity of their child's disability. More recent studies, designed specifically to explore positive perceptions in families, have also identified the positive impact that the child may have on the family generally. Studies have identified dimensions that may be specific to raising a child with a disability, such as: source of joy and happiness, personal growth and strength, increased family closeness, increased sensitivity to others, and opportunities to expand one's social and political activities and contacts (Stainton & Besser, 1998; Scorgie, Wilgosh & McDonald, 1999; Scorgie & Sobsey, 2000).

More recently research approaches have tended to be quantitative in nature and focus on identifying predictor variables for parental positive perceptions, and on the relationship between positive perceptions and parental coping. Findings suggest that that mothers' perceptions of the positive impact of the child and its effect on the family are positively predicted by the use of reframing coping strategies, the helpfulness and usefulness of support from family and friends, and the caregiving demand (Hastings *et al*, 2002) and by parental confidence and sense of coherence (Mak, Ho & Law, 2007). In a comprehensive review of the literature Hastings and Taunt (2002) identified a number of key issues related to research on the positive perceptions and experiences of families of children with disabilities. These include: family members do report a range of positive perceptions and experiences; the presence of positive perceptions and experiences seems to occur in concert with negative or stressful experiences; although typically reporting more stress than do families of children without disabilities, families of children with disabilities do not seem to report fewer positive perceptions; positive and negative perceptions seem to be predicted by different factors. They

suggest that positive perceptions may serve some function for families of children with disabilities as a means of coping with the experience of raising such a child.

In summary, there is considerable evidence to suggest that parents perceive positive impacts from caring for a child with an intellectual disability. Much of this is anecdotal evidence from studies which were not specifically designed to elicit positive perceptions. Studies which focus specifically on positive parental perceptions have tended to be either qualitative in nature, or limited by small samples which have been purposefully selected. While all of these studies included parents of children with a variety of age ranges and disability severity to ensure comprehensive representation, this coupled with small sample sizes, makes it impossible to look at differences in positive perception related to disability severity or the existence of co-morbid conditions. Consequently the generalisability of the findings of these studies to the population of interest in the current study is not clear. Studies specifically focusing on positive parental perceptions of their children with complex or life-limiting disability are absent from the research literature which makes it difficult to draw conclusions regarding parental perceptions in relation to young children with life-limiting neurodevelopmental disabilities. It is also worth noting that, despite the focus on parents' positive perceptions, participants in all studies commented on aspects of burden and some element of negative interaction with services, suggesting the concepts of positive perceptions and burden are not bipolar positions and but rather occur concurrently.

3.7.2 The Experience of Parenting a Child with a Disability

An additional feature of the research literature related to children with disabilities is the general considerations of the meaning of parenting such children. This not an area that has received much attention in the context of childhood life-limited illness, although it received a cursory mention in a study of parents of children with life-limiting illnesses by Steele and Davies (2006), and is a feature of parents' descriptions of caring for their technology-dependent child in a study by Carnevale *et al* (2006).

By their nature studies exploring the meaning of parenting a child with a disability are qualitative in design. The findings of these studies are consistent and present a parenting experience generally characterised by a constant tension between concurrent positive and

negative emotional states and appraisals which evolve and change over the course of the parenting experience (Kearney & Griffin, 2001; Green, 2007a; Hauskov-Graungaard *et al*, 2011). In a phenomenological study of six parents focused on parenting a child with various levels of developmental disability Kearney and Griffin (2001) describe a model characterised by tensions between states of joy and sorrow which explain parents' concurrent contradictory emotions and thoughts. In this study parents' attributed states of joy to their child, while those of sorrow were attributed to parents' dealings with other people's frequent messages of negativity and hopelessness. Parents in this study spoke of: confusing and conflicting emotions; ambiguous prognoses; conflicting perceptions between themselves and health professionals; of not knowing what to expect and sometimes simply not knowing what to do. The relevance of this study to the population of the current study is limited by the fact that two of the children in Kearney and Griffin's study (2001) had an acquired developmental disability and so would have been parented in an entirely different context prior to the occasion of their disability. In addition, the study included a wider range of levels of developmental disability. Conversely Green (2007a) describes the mothers of the disabled children in her mixed methods study as "tired not sad". The mothers who participated in this study reported significantly higher levels of objective than subjective burden demonstrating that mothers perceived parenting a child with a disability in terms of socio-cultural constraints rather than emotional distress. The fact that this study included both physically and intellectually disabled children may account for the difference in the findings.

Hauskov-Graungaard *et al* (2011) provide a temporal dimension on the experience of parenting a child with a severe disability. In a longitudinal grounded theory study of parental emotions and coping they suggest that parents continuously created and sustained personal resources through a process of cognitive positive reappraisal of the present conditions, and the consequences of these conditions. The authors label this process "resource-creation" and found it to be an integrated part of the continuing process of coping process for parents throughout the course of the study. Concomitantly with resource-creation parents in this study also experienced what the authors describe as "resource-deterioration", either as a result of their own despair and fear, or the influence of external stress factors such as exhaustion or non-empathetic health professionals.

In summary, because of the nature of the phenomena being investigated, research into the experience of life as a parent of a child with disability are small scale qualitative studies using purposefully selected samples. These studies have often included a wide age-range of children amongst their samples, and included children a broad range of both physical and intellectual disabilities, some of which were acquired and some congenital. In this context the degree to which the findings can be generalised specifically to young children with life-limiting neurodevelopmental disabilities is not clear.

3.7.3 Summary of Research Literature Related Meaning and Appraisal

Meaning and appraisal are considered bridging concepts between family coping and family adaptation in Family Stress, Adjustment and Adaptation Theory (McCubbin *et al*, 1983). Although this is not an aspect of childhood life-limiting illness that has been widely explored it had received much attention on the literature related to childhood disability. Findings suggest that parents frequently describe positive perceptions of their children with disabilities in terms of the contribution the child makes both to personal growth and to family life, and that the experience of parenting a child with a disability is characterised by state of tension between concurrent positive and negative emotional states and appraisals.

3.8 Synopsis of Research Literature Review

Children with life limiting conditions, regardless of the nature or origin of the condition, have wide-ranging, ongoing and often complex needs which place a considerable practical, emotional and social burden to their families and carers. Although rarely specified as a particular and cohesive group, evidence seems to suggest suggests that families of children with life-limiting disabilities are subject to the same burden of care and family impact. Providing care for these children has implications beyond the child extending to all members of the family and all aspects of family life and functioning. The burden on parents, principally mothers, of caring for such children is widely reported in published literature with studies reporting the time and financial costs, the physical and emotional burden of care provision and the logistical complexities of trying to provide and organise care for a child with a complex condition. However it is also reported that the experience of care provision

can have a positive impact on the family. In this context parents have described broadened horizons, increased awareness of inner strength and expanded social and community networks.

Sibling literature reports that siblings of children with a chronic illness experience some negative effects compared to those who do not have a sibling with chronic illness. It has also been demonstrated that illnesses which necessitate daily treatment regimes are more strongly associated with negative effects on siblings than those that do not. Most young children with life-limiting neurodevelopmental disabilities require daily treatment of some kind and the potential for adverse effects on siblings is therefore highly relevant to this group. Concerns about adverse effects on healthy siblings can add to parental stress.

Despite the primary care focus of healthcare policy, and the desire of mothers to care for their child at home, a growing body of research indicates that the healthcare system has been consistently failing to adequately meet the needs of such children and their families. Consistent themes in the literature related to service provision to this group of children and their families include: access difficulties, a lack of uniformity in all aspects of service provision and quality; bureaucracy; and a lack of family-centred care. Strickland *et al* (2004) suggest that these essential characteristics of services remain problematic for all children with special health care needs. Many families of children with life-limiting conditions and complex disabilities describe how in addition to the time and effort required to care for their child they expend considerable time, energy, and financial resources on advocacy and other activities due to a poorly coordinated and often unresponsive health system of service delivery.

3.9 Limitations and Gaps in the Literature

It is acknowledged that an overlap exists between the care required by children with life-limiting illnesses and that required by children with life-limiting disabilities (DOH&C, 2005, 2010). The precise extent of this overlap is not known however, as research studies focusing specifically on young children with life-limiting neurodevelopmental disabilities and their families are largely absent. Consequently the specific needs of these children and their

families have become invisible and indistinguishable from the needs and experiences of children and families in the samples into which they are subsumed.

In the research literature children with life-limiting neurodevelopmental disabilities have crossed the borders of studies exploring childhood chronic conditions, childhood life-limiting and life-threatening illnesses, and childhood disability. While they potentially share features with all of these children they also have additional unique features which they do not share. Similarly while they share essential features with other children with neurodevelopmental disabilities, the complexity of their conditions and limited prognosis marks them as unique from other neurodevelopmentally disabled children. As a result the extent to which current research findings can be applied to this specific group of children is not clear.

There is scant information of the morbidity experienced by children with life-limiting conditions generally, and very few studies that have explored issues related to this population of children and their families as a unique group. Subsequently the conclusions that can be drawn about the specific needs of these children and their families are limited, and in the absence of empirical evidence remain at best a guess. Consequently important questions that are integral to the development of policy solutions remain unanswered in the research literature. Specifically these gaps in the literature include: what is the demographic and health status profile of young children with life-limiting neurodevelopmental disabilities? What services do these children and their families require, and what access barriers do they face? How effective are the services that these children and families receive? What can be done to improve the health care system and delivery of services to this group of children and their families to best meet their needs? It is the aim of this research study to address these deficits and to accurately define the needs and experiences of this group of children and their families as a distinct and individual group.

3.10 Conclusion

Caring for a child with any health-related problem entails greater than average time demands, care-giver burden, financial costs and employment constraints. Research suggest that in the context of childhood life-limiting illness, or for children dependent on medical technology, these additional demands are considerable, and can significantly affect the physical and

psychological health of care-givers and family functioning. The degree to which these negative impacts are caused entirely by the additional demands of caring for a child with complex medical needs is hard to say as the literature is generally consistent with a stress-process model and identifies several intervening factors which act to mediate the impact.

This chapter has presented a critical review of the literature related to, but not specifically confined to, the population of this study. The difficulties of terminology and definitions have been identified, and the impact of these ambiguities on the review of the literature has been discussed. There are still areas which need to be explored in relation to this specific population of children and their families, and these gaps in the literature have been identified, and have been related to the research questions of this study.

Chapter 4: Overview of the Present Study

“Would you tell me please which way I ought to go from here?” asked Alice.
“That depends a good deal on where you want to get to”, said the cat.

Lewis Carroll (2007:58) *Alice’s Adventures in Wonderland*.
Penguin Popular Classics

4.1 Introduction

The purpose of this chapter is to provide an overview of the current study and the methodological approach and design used, namely a mixed methods design. The study proceeds in two phases. Phase One uses a sequential explanatory design, and Phase Two a Delphi design. The chapter begins by presenting the aims and objectives of the study and the research questions to be addressed. Next, an overview of the study in its entirety is presented (the specific methodological aspects of the different phases and stages are presented in greater detail in Chapters five to seven which deal individually with each phase). The relationship between the research questions and the study methodology is explored and the manner in which the different phases of the study relate, both to each other and to the research questions, is described.

The chapter concludes with a discussion of the ethical principles underpinning the study as a whole, and the application of these principles to each phase of the study.

4.2 Aims and Objectives of the Study

The overall intention of this research is to provide a detailed and reliable evidence base that relates to the current health system as it pertains to young children with life-limiting neurodevelopmental disabilities and their families. Research in this area is needed to provide the premise of fact upon which any national policy must be built, and is congruent with the children’s palliative care research agenda proposed by Quinn *et al* (2005) and Steele *et al* (2008) who identify the need to explore the specific palliative care needs of individual populations of childhood conditions.

The study proceeds in two phases. Phase One explores the specific palliative care needs of young children with life-limiting neurodevelopmental disabilities and their families. This phase of the study focuses on the family's own perspective and has three main objectives -

- To explore the palliative care needs of children with life limiting neurodevelopmental disability and their families.
- To explore the impact of providing care to children with life limiting neurodevelopmental disability on the family
- To explore families experiences of the services available to them.

Phase Two of the study relates to the current provision of services to this population of children and their families. This second stage focuses on service provider's perspectives, and has one main objective which is –

- To explore health care providers perceptions of the services currently available to young children with life-limiting neurodevelopmental disabilities and their families.

4.3 Research Questions.

The specific research questions to be addressed by the study are –

1. What challenges do children with life-limiting neurodevelopmental conditions and their families commonly experience?
2. What are the palliative care needs amongst this group of children?
3. What is the level of psychological distress among parents of children with life-limiting neurodevelopmental conditions and what resources and coping mechanisms are used?
4. What impact does caring for a child with a life-limiting neurodevelopmental condition have on the family?
5. What level of social support is available to these parents?
6. What specific factors exacerbate or ameliorate the negative impact of care provision on families?
7. Can the variables that have the most significant impact on the family be identified?

8. What services are available to these children and their families, and how are these services perceived?
9. What are the agreed goals of care, from a services perspective, for children with life-limiting neurodevelopmental condition?
10. How well do current services function to meet the needs of children and their families?
11. What changes are necessary to improve the care of children with life-limiting neurodevelopmental conditions?

Research questions one to eight focuses specifically on the child and family and relate to the aims of the first phase of the study. Research questions nine to eleven focus on the perspectives of service providers, and relate to the study's second phase. The research questions are grouped according to their particular phase and aim in Figure 4.1.

Figure 4.1 *Relationship of Research Questions to Study's Phases and Aims*

<p><i>Phase One</i> AIM</p> <p>(1) To explore the palliative care needs of children with life limiting neurodevelopmental disability and their families.</p> <p>(2) To explore the impact of providing ongoing care to children with life limiting neurodevelopmental disability on the family</p> <p>(3) To explore families experiences of the services available to them.</p>	<p><i>Research Questions</i></p> <p>(1)What challenges do children with life-limiting neurodevelopmental conditions and their families commonly experience?</p> <p>(2) What are the palliative care needs amongst this group of children?</p> <p>(3) What is the level of psychological distress among parents of children with life-limiting neurodevelopmental conditions and what resources and coping mechanisms are used?</p> <p>(4) What impact does caring for a child with a life-limiting neurodevelopmental condition have on the family?</p> <p>(5) What level of social support is available to these parents?</p> <p>(6) What specific factors exacerbate or ameliorate the negative impact of care provision on families?</p> <p>(7) Can the variables that have the most significant impact on the family be identified?</p> <p>(8) What services are available to these children and their families, and how are these services perceived?</p>
<p><i>Phase Two</i> AIM</p> <p>(1) To explore health care providers perceptions of the services currently available to young children with life-limiting neurodevelopmental disabilities and their families.</p>	<p><i>Research Questions</i></p> <p>(9) What are the agreed goals of care, from a services perspective, for children with life-limiting neurodevelopmental condition?</p> <p>(10) How well do current services function to meet the needs of children and their families?</p> <p>(11) What changes are necessary to improve the care of children with life-limiting neurodevelopmental conditions?</p>

4.4 Choice of Mixed Methods Design

The overall objectives of the study could best be achieved by adopting a dual focus which comprehensively considered the perspectives and experiences of both parents and healthcare providers. In this respect the study needed to acquire information that was both circumstantial and experiential, and situated both at the level of service users and service providers.

Additionally, in order that a comprehensive exploration could be undertaken, the study required the collection of both quantitative and qualitative data. No single research method or design could be found which would comprehensively address all of the study's research questions, consequently a mixed methods approach was chosen on the basis that each method functions as a tool that is calibrated to answer specific research questions, but not others. Gilbert (2005) suggests that while quantitative and qualitative research procedures can provide different level perspectives on the social world, in combination they can be used to explore different dimensions of a problem. In a mixed methods approach it is the research question rather than an ideological commitment to any particular kind of methodology that becomes central to the study (Bryman, 2007). Greene (2007:118) contends that "the greatest potential of mixed methods inquiry is the generative possibilities that accompany the mixing of different ways of knowing, perceiving and understanding".

Each method used in this study was chosen on the basis of its ability to address particular research questions and aspects of the study. Consequently confining the study to only one method would have severely curtailed its ability to comprehensively address all of the questions. The use of a mixed methods approach is a unique contribution of this study in the context of previous research in this area. By using mixed methods, the analysis can contribute to an increasingly three-dimensional picture of the palliative care needs and service delivery to young children with life-limiting neurodevelopmental disabilities and their families.

4.5 The Mixed Methods Design of the Current Study

As previously outlined, this study proceeded in two phases. Phase One was concerned with the young child with a life-limiting neurodevelopmental disability and their family and collected data from parents using both a postal survey (Stage One) and interviews (Stage Two). This phase employed a sequential explanatory mixed methods design.

Phase Two of the study was concerned with the perspective of service providers. This second phase utilised a Delphi design, and involved the collection of both qualitative and quantitative data from an expert panel of health service providers. The following section provides an overview of the design for each phase. These are subsequently described in detail in Chapters five to seven.

4.5.1 Phase One: Sequential Explanatory Mixed Methods Design

Sequential mixed methods designs involve multiple phases of data collection in which the research purpose, and particular set of research questions, determine the particular sequence (Andrew & Halcomb, 2009). Sequential designs may be either explanatory, in which the quantitative data is collected first followed by the qualitative element of the study, or exploratory, in which the qualitative data is collected first and followed by the quantitative element of the study (Creswell & Plano-Clark, 2007). An additional salient consideration in a mixed methods design is the weight, or priority, given to the data elements within the study which determines the study's theoretical drive (Morse, 2003). Andrew and Halcomb (2009) suggest that in explanatory designs the weight is usually, but not always, afforded to the quantitative element of the study, while exploratory designs usually, but not always, affords the weight to the qualitative element of the study.

A sequential explanatory design was chosen for the implementation of Phase One as the purpose of the qualitative data collected was to help explain, and expand upon, the findings of the initial quantitative round. An exploratory design is generally considered to be more appropriate to studies in which little is known about the subject area (Creswell & Plano Clarke, 2007; Andrew & Halcomb, 2009).

The goal of mixed methods research is to draw on the strengths and minimise the weaknesses of both quantitative and qualitative approaches in a single study or across studies (Teddle & Tashakkori, 2003, 2009), with the methods chosen on the basis of complementary strengths and non-overlapping weaknesses (Johnson & Onwuegbuzie, 2004). In respect of this study the structured data collection techniques of the survey, with its standardised measurement scales, brought potential strengths of quantification, precision, and reliability. Alternatively the qualitative data from parents' interviews brought a fuller, more holistic understanding of

the experience of caring for a young child with a life-limiting neurodevelopmental disability by allowing parents their own particular perspectives and concerns in the context of their own experience. This is consistent with an integrated sampling strategy, and with the use of an explanatory mixed methods design in which the quantitative data and their subsequent analysis provide a general understanding of the research problem, while the qualitative data and their analysis refine and explain the statistical results by exploring participants in greater depth (Tashakkori & Teddlie, 1998; Creswell & Plano Clarke, 2007; Creswell, 2008).

Phase One of the study represents a fully mixed design in that integration occurs across all strands of the study (Onwuegbuzie & Johnson, 2004). The research questions are descriptive, predictive and exploratory, representing both quantitative and qualitative approaches, with further integration occurring at both the sampling and data interpretation stages. Survey data was analysed and parents for interview purposefully selected from the quantitative sample on the basis of their Impact on Family scores (described in Chapter six). This allowed representation of a variety of experiences and ensured that anomalies in the data could be explained or explored in greater depth. Integration also occurred at the data analysis stage whereby the two stages of the phase were brought together and areas of convergence and divergence explored [Figure 4.2]. Thus the qualitative data provided an explanation for, and augmented, particular parts of the quantitative study in order to further enhance and add depth to the quantitative data in line with the overall objectives of a sequential explanatory design.

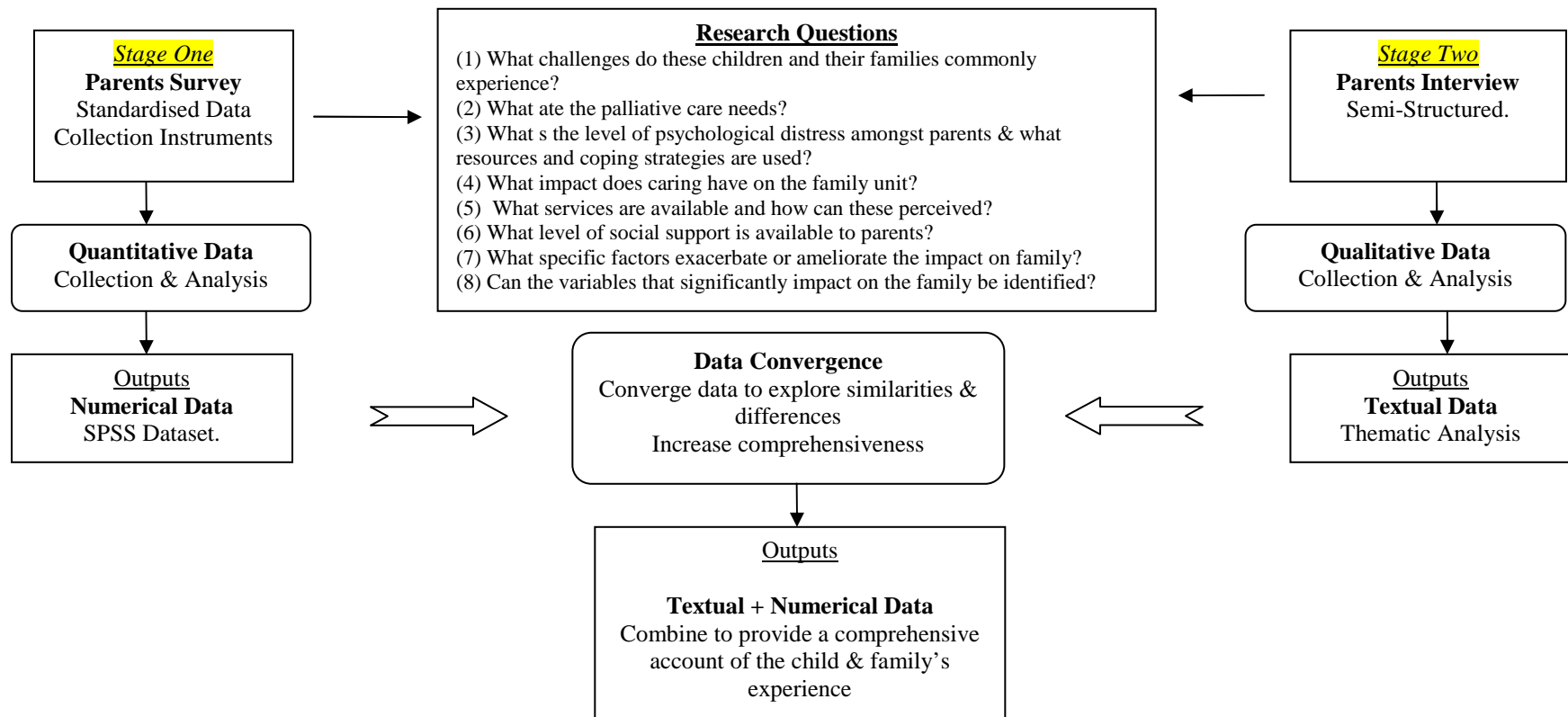
Figure 4.2 *Sequential Explanatory Mixed Methods Design for Phase One*

Aim: To explore the palliative care needs of young children with life-limiting neurodevelopmental disabilities and their families.

To explore the impact of providing ongoing care on the family.

To explore families experiences of the services available to them.

Sample: Young Children with Life-Limiting Neurodevelopmental Disabilities and their Families



4.5.2 Phase Two: Delphi Design

Linstone & Turoff (1975:3) describe Delphi as “a method for structuring a group communication process so the process is effective in allowing a group of individuals, as a whole, to deal with a complex problem”. Delphi was chosen for the second phase of the study as the method is consistent with the aim of exploring expert opinion. It has been widely used as a constructive method in facilitating controlled, rationale group communication to develop knowledge for decision making (Du Plessis & Human, 2007).

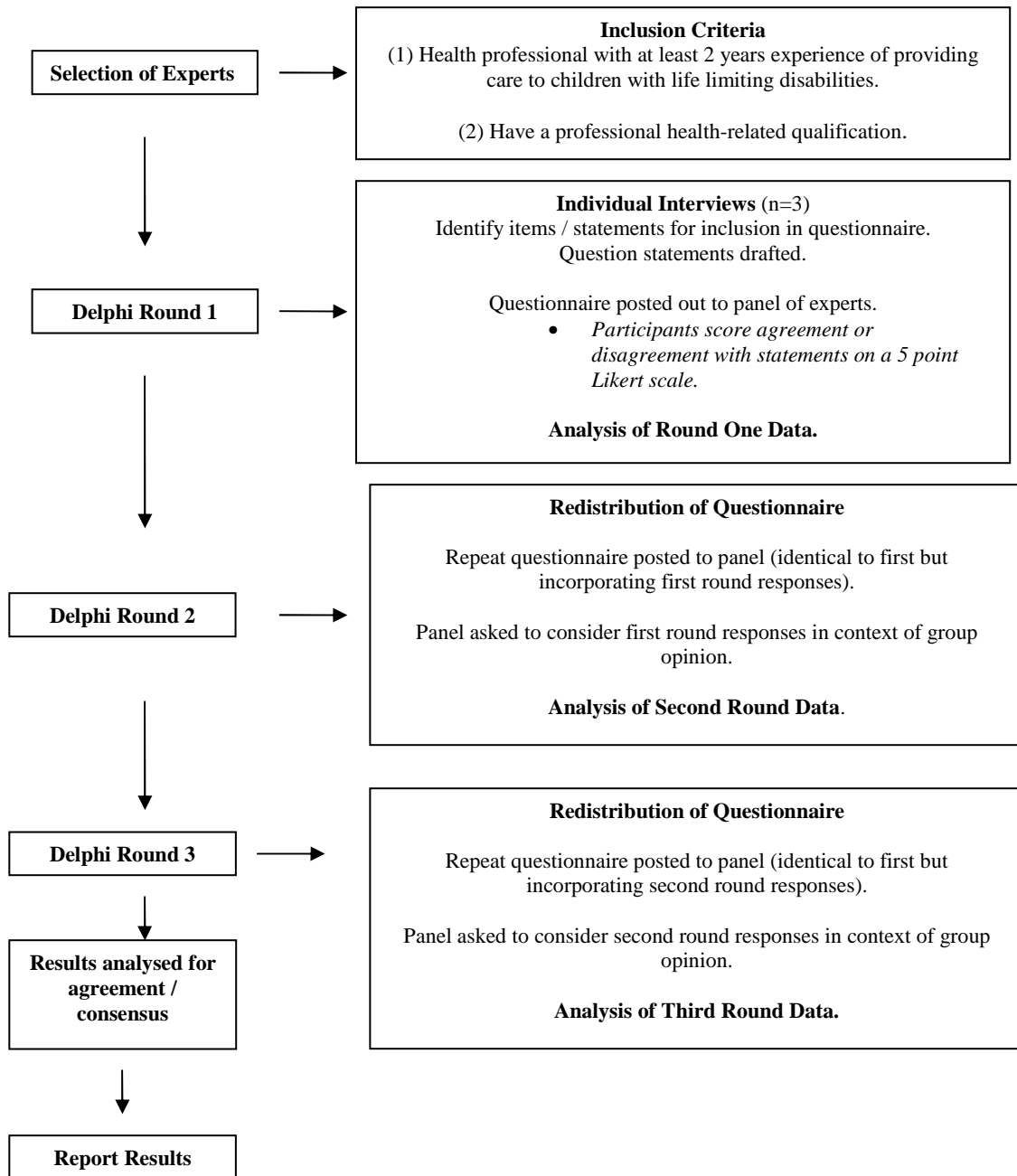
Although widespread use of Delphi has resulted in various approaches, all applications describe common features of the technique, namely that it involves a series of sequential questionnaires or rounds, interspersed by controlled feedback, designed to elicit the most reliable consensus of opinion amongst a panel of experts (Turoff, 1975; Dalkey, 1969; Meade & Moseley, 2001; Du Plessis & Human, 2007). The technique is based upon the assumption that combined numerical estimates of participants’ views would, in general, lead to more reliable estimates than estimates from a single individual (Murphy *et al*, 1998), or as Dalkey (1969:v) succinctly puts it “based upon the premise that two heads are better than one”. The Delphi process followed in this study is outlined in Figure 4.3. The phase is described in detail in Chapter Seven.

It is not usual to see Delphi incorporated into a mixed methods study, nor is it identified as part of any traditional typology mixed methods designs. However, it may be argued that Delphi itself contains many of the essential features of a sequential exploratory mixed methods approach particularly where, as often is the case, qualitative data is collected at the outset and forms the basis of the quantitative questionnaire used in the iterative rounds as is the case in this study.

Figure 4.3 Delphi Design for Phase Two

Research Questions

1. What are goals of care for children with life-limiting neurodevelopmental disabilities?
2. How do current services achieve these goals?
3. What changes are necessary to current services to improve the care of children with life-limiting developmental disabilities?



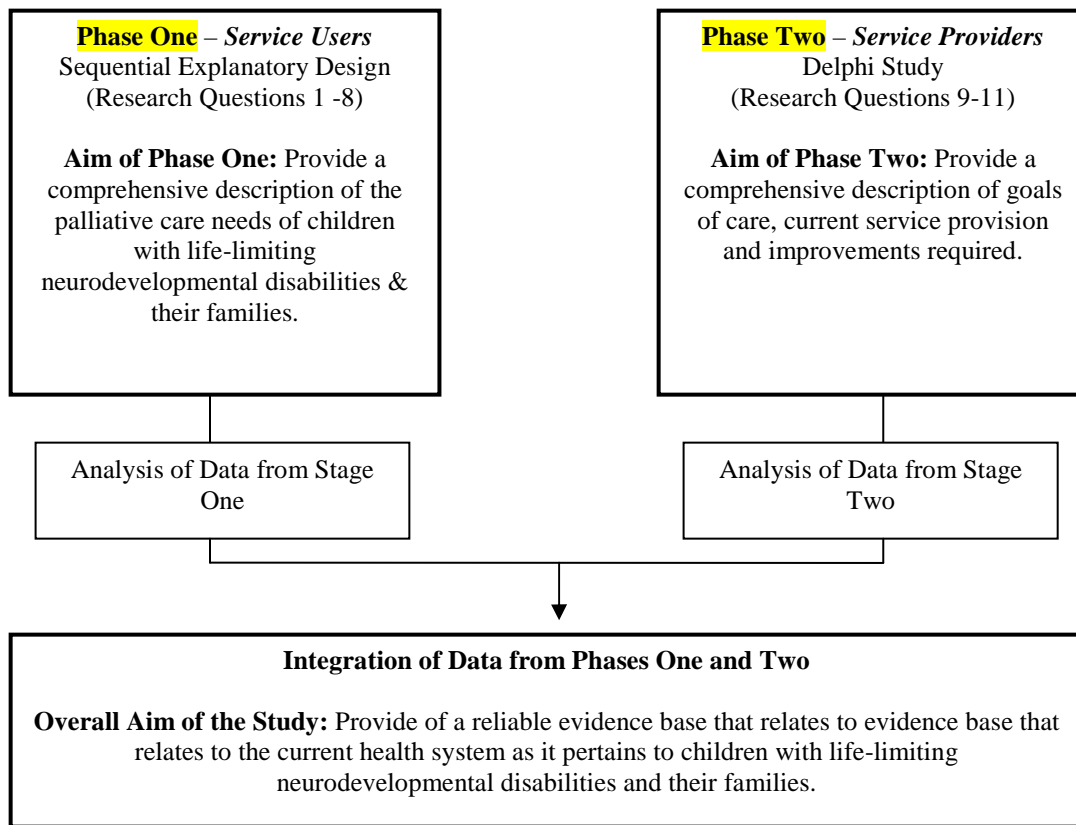
Adapted from Jones & Hunter 1995

4.6 Integration of the Study Phases

Integration is a critical feature of any mixed methods study (Johnson & Christensen, 2004; Johnson *et al*, 2007). Onwuegbuzie and Johnson (2004) propose that essentially any research design occupies a place on a continuum from not mixed to fully mixed, with the exclusive use of either a quantitative or qualitative approach occupying one end of this continuum and fully mixed methods the other. They suggest that fully mixed methods designs represent the highest degree of mixing research methods and research paradigm characteristics. Fully mixed designs involve using both qualitative and quantitative research within, or across, one or more of four components in a single research study: (a) the research objective; (b) the type of data and operations; (c) type of analysis; and (d) type of inference. Alternatively in partially mixed methods, the quantitative and qualitative phases are not mixed within or across stages. Instead both elements are conducted either concurrently or sequentially in their entirety before being mixed at the data interpretation stage. The integration of Phase One has already been described and involves integration across all four components of the study, which constitutes a full mixed methods design according to Onwuegbuzie and Johnson's criteria (2004).

Phase Two is not fully integrated with Phase One since the two phases address different research questions, and different samples are used. Despite this, the phases are integrated at the level of the overall aim of the study (described in Chapter One), and the findings of both phases are integrated in Chapter Eight in order that overall conclusions can be reached and recommendations made. In this respect the study overall constitutes a partially mixed methods design. The integration of the different phases of the study is presented in Figure 4.4.

Figure 4.4 *Integration of Study Phases*



The previous sections of this chapter have described the overarching mixed methods design of this study. In order to achieve its aim and answer its research questions the study proceeds in separate phases. However, the overarching design of the study was informed by certain ethical principles which guided its conduct through both phases. The remainder of the chapter discusses these principles and their application to the overall study.

4.7 Ethical Issues in the Design of the Study

At a general level this study deals with areas which have been designated as sensitive topics in healthcare. Lee (1993:4) defines sensitive research as “research which potentially poses a substantial threat to those who are, or have been, involved”. Sensitive contexts pose particular pragmatic challenges for researchers, and in such contexts there is an increased onus on researchers to be aware of their ethical responsibility to research participants (Lee,

1993; Renzetti & Lee, 1993). In addition, the participants in this study fall into a category designated as a “vulnerable” research population (Link & Phelan, 1996; Guralnik & Leveille, 1997) which suggests that they are at greater risk of harm or exploitation if sound ethical principles are not adhered to. Finally, it has been suggested that poorly designed research is inherently unethical since it wastes subjects’ time and energy if the results are less than useful (Iphofen, 2009; Israel & Hay, 2009; Strohm Kitchener & Anderson, 2011).

This research proposal was submitted for review of its scientific merit and ethical acceptability to the Ethics Committee of Dublin City University. It was submitted in three separate parts (independently for parents’ surveys, parents’ interviews and Delphi) with ethical approval obtained from the committee in advance of proceeding with each phase. Permission for use of the quantitative data collection instruments was obtained from the individual, or institution, holding the copyright for the particular scale. This was not required for the General Health Questionnaire-28 [GHQ-28] as it is publicly available, and was purchased for use in this study, from GL Assessment Ltd. Chiswick High Road, London.

Essentially most ethical guidelines for conducting research are based upon the principle of respect for participants’ human rights, and adopt the position that research involving human subjects must not violate any universally applicable ethical standards (World Medical Assembly [WMA], 1964; Council for International Organisations of Medical Sciences [CIOMS] & WHO, 1993). In order to comply with this position certain ethical principles were used as a framework to guide the researcher and the research process. This framework included the principles of respect for the person; beneficence and nonmaleficence; and fidelity and justice ((Beauchamp & Childress, 2001; Strohm Kitchener & Kitchener, 2009). The application of each of these principles to the conduct of the study is discussed below.

4.7.1 Respect for Persons

The fundamental ethical consideration incorporated in the principle of respect for persons relates to the issue of personal autonomy (Beauchamp & Childress, 2001; Strohm Kitchener & Kitchener, 2009). This principle requires that those who are capable of deliberation about their personal choices should be treated with respect for their capacity for self-determination, and that persons with impaired or diminished capability should be protected. Autonomous

individuals make decisions that are “free from both controlling interference by others and personal limitations, such as inadequate understanding, that prevent meaningful choice” (Beauchamp & Childress, 2001:58). In this context it is required that researchers obtain “informed consent” from participants before conducting a research project. Israel and Hay (2009) suggest that the concept of informed consent implies two related activities: participants’ first need to comprehend, and second to agree voluntarily to, the nature of the research and their role within it. Consequently informed consent places an obligation on the researcher to provide potential participants with full, relevant information on the research project, and can be considered voluntary only in the absence of actual or implied coercion of participants.

4.7.1.1 Autonomy & Informed Consent of Participants in Phase One

Before making a decision about whether or not to participate in this study all potential participants were issued with an Introductory Letter [Appendix B] and comprehensive Information Booklet [Appendix C] which provided full details of the study including the requirements of participants should they agree to be involved. The booklet also included a contact telephone number and email address for the researcher. Participants were encouraged to contact the researcher if they required any additional information or wanted to discuss any aspect of the study before making a decision about whether or not to participate. All of the documents intended for participants (letters, booklet and consent forms) were reviewed by the National Adult Literacy Association [NALA] and awarded the Plain English Mark. This ensured that they were as comprehensible and accessible as possible to all individuals.

Given the sensitive nature of the study questionnaires did not accompany the Information Booklet for parents. Parents, who after reading the information booklet returned a completed consent form [Appendix D], received a copy of the study questionnaire by mail [Appendix E]. The voluntary nature of participation was emphasised in all correspondence, as was each participant’s right to withdraw from the study at any time without explanation or penalty.

Information about the interview stage of the study was included with the letter accompanying the postal questionnaire [Appendix F]. This simply informed participants that the researcher was also seeking individuals who would be willing to discuss their experience of caring for

their child and the services available to them. No further information was included at this stage for fear of overburdening participants and causing confusion about what was being consented to. Instead participants were asked to return a second consent form if they were willing to be contacted by phone to discuss participation in the interview stage [Appendix G]. It was emphasised that this consent form indicated only that they were willing to be contacted to discuss potential participation, and was not consent to be interviewed.

If participants agreed, and were selected for interview (as outlined in Chapter 6), they were telephoned at a time they had specified as convenient. During this telephone conversation participants were informed of the purpose of the interview, the topics to be discussed, and the process of conducting the interview. Any questions participants had were addressed. Verbal consent to be interviewed was obtained over the phone, and a convenient time and place arranged to conduct the interview. Participants then received a confirmatory letter which reiterated the information already provided, and included a telephone number on which they could contact the researcher if they changed their mind about being interviewed. Written consent was obtained from all interviewees in advance of conducting the interview [Appendix H].

Participants for this study were recruited through the Jack and Jill Children's Foundation charity (described in Chapter Five). Initial information was posted directly to potential participants through the Foundation. It is suggested that the use of such sampling frames may provoke anger and anxiety in potential participants, and that recruitment of participants through such an agency may influence the tone of responses (Lee, 1993; Renzetti & Lee, 1993). However great care was taken to emphasise to participants that the research project was being conducted independently of the Foundation and that the Foundation had no involvement in the study beyond agreeing to the use of their database as a sampling frame. This point was accentuated in the information booklet, the letter accompanying the survey, and again in advance of the interviews with participants.

In addition, it was emphasised that participation in the study was entirely voluntary. Participants were reassured at every stage that there would be no negative consequences for

those who chose not to participate, or for those who decided to withdraw from the study before its completion.

4.7.1.2 Autonomy & Informed Consent of Participants in Phase Two

Participants for the Delphi expert panel were recruited through a combination of purposeful and snowball sampling (discussed in detail in Chapter Seven). Unlike Phase One participants this was not considered a vulnerable research population, nor was the topic of investigation a particularly sensitive one.

Potential participants received a letter outlining the details of the study and inviting participation [Appendices I & J]. A contact number for the researcher was included should any additional information be required. Those who agreed to participate by returning a completed consent form [Appendix K] received instructions and a postal questionnaire for Round One [Appendices L & M]. The recruitment of participants to both phases of the study is discussed in detail in subsequent chapters of the thesis.

4.7.2 Beneficence and Nonmaleficence

The concepts of beneficence and nonmaleficence are often interlinked and refer to researchers' ethical obligation to maximise benefits and minimise harms to research participants (Israel & Hay, 2009). In an attempt to balance the risk of harm against the potential benefits that may accrue from participation researchers need to consider several factors: these include the particular kinds of harm that may occur; how likely these are to occur; the ways in which these harms can be minimized; and the ways of maximizing both short and long term benefits (WMA, 1964). It is generally accepted that risks and benefits should be balanced and shown to be in a favorable ratio (Beauchamp & Childress, 2001; Strohm Kitchener & Kitchener, 2009).

While some purely scientific research may have the potential to cause physical harm to participants, in social science research the concept of harm is generally most likely to involve psychological distress, discomfort, social disadvantage, or invasion of privacy (Israel & Hay, 2009). This study deals with sensitive areas which are private to participants and may be

emotionally charged. Subsequently either, or both, of the two stages of Phase One (quantitative and qualitative) could potentially pose a psychological or intrusive threat to participants. In this respect a plan for the support and protection of participants was developed at the outset of the study.

4.7.2.1 Protection of Study Participants.

Because of the nature of the phenomena being investigated it was possible that the GHQ-28, used as part of the survey questionnaire, would identify participants suffering from high levels of psychological distress. It was planned that, should this occur, the identity of the participant would be traced using the unique identification code assigned to individual questionnaires. The researcher would then contact the individual by phone to assess the situation. If necessary permission would be sought from the participant to contact the appropriate primary care services for additional support. It was not necessary to enact this plan during the study.

The researcher who conducted the qualitative interviews had extensive experience of interviewing and supporting vulnerable families and individuals. She was experienced in dealing with the emotive issues that can arise during home interviews. On conclusion of each interview the participant was given contact details for the researcher should they wish to later discuss any aspect of the interview or the interview process. Where interviewees became distressed during the interview (as happened in three cases) they were offered the option of discontinuing the interview or of taking a break. All declined and wished to proceed. After a difficult interview, the researcher remained with, and provided support to, the interviewee as needed. A follow-up telephone call was made two days later to check how the individual was doing, provide additional support, or refer the participant to their primary care services if appropriate and agreeable. No participant required referral to additional services.

No plan for the support of participants in the Delphi phase of the study was developed. Participants in this phase were expert health professionals giving their opinion of service provision. This stage of the study was low risk and no harm to participants was anticipated.

4.7.2.2 Maximising Benefits for Study Participants.

The concept of beneficence is more elusive in social science research and Israel and Hay (2009) suggest that researchers have tended to concentrate on acting to avoid causing harm to participants rather than acting to benefit them. There were no direct benefits to participants for being involved in this study beyond providing them with a platform and an opportunity to express their opinion and experiences. However, Iphofen (2009) suggests that to be beneficence can entail a wide range of actions including improving understanding of the phenomena, or providing indirect benefits in the form of social policy in the topic area. The overall objective of this study is compatible with this broader view of the concept of beneficence.

4.7.3 Fidelity & Justice

In general, the principle of Justice refers to the ethical obligation to treat each person in accordance with what is morally right and proper, and to give to each person what is due them. In social science research however it is less to do with the relationship between the researcher and participant and more to do with the equitable distribution of both the burdens and benefits of research participation (Strohm Kitchener & Kitchener, 2009). In contrast, the principle of Fidelity is at the core of the relationship between social science researchers and research participants and involves the concept of trust. Beauchamp and Childress (2001:312) propose that fidelity “justifies the obligation to act in good faith, to keep vows and promises; fulfill agreements and discharge fiduciary responsibilities”. The concept of fidelity commonly implies two activities. One of these is the obligation of veracity (Beauchamp & Childress, 2001), the second the promise of confidentiality (Strohm Kitchener & Kitchener, 2009).

Beauchamp and Childress (2001:284) equate veracity with “comprehensive, accurate and objective transmission of information”. It involves the concepts of truth and the absence of deception. Comprehensive and truthful information was provided to all participants in this study, in a timely manner, and an accessible format. All of the study participants had both telephone and email contact details for the researcher, and any questions or requests for information were promptly and respectfully responded to. The study involved no deception of participants. Participants were informed that, once completed, it was intended to publish the

study findings in a peer-reviewed journal and present the findings at a conference. Participants were reassured on the measures taken to preserve their anonymity and no participant objected to this use of their data. All participants were also given the option of receiving a copy of the findings.

4.7.3.1 Privacy & Confidentiality of Participants

Anonymity was possible at varying levels depending upon phase of the study. Several factors impacted upon this. In the context of parents this included the relatively small population from which the sample of parents was drawn, and the sampling frame used to identify participants. In the context of the Delphi it again included the small and elite number of experts from which to draw the sample, and the snowball method of sampling used to identify the expert panel. Despite this, every effort was made by the researcher to remove, as far as possible, the opportunities for others to deduce participants identity from the compiled data.

Acknowledged and justifiable exceptions exist to the kinds of information that can be considered confidential in policy and practice (Beauchamp & Childress, 2001). Before consenting to participate in the study all participants were informed that confidentiality would be broken in the case of a concern about the safety of a child in accordance with the researcher's legal responsibility (Government of Ireland, 1991). In such a case the researcher is obliged to bring this to the attention of the primary service provider in order that the case can be followed up. This was not an issue during the study. The steps taken to ensure participants privacy and confidentiality will now be discussed independently for each phase of the study.

Completed parents survey questionnaires contained no information by which participants could be identified beyond a unique identification code which allowed identification by the researcher. This code was required for two reasons: firstly to allow the identification of individuals who may have required additional support as outlined above; secondly to allow data from the questionnaires to be matched with interview data. All data returned in the questionnaires were treated as confidential. Data from paper questionnaires were transferred into a secure, password protected computer programme, which was in the sole custody of the

researcher throughout the study. When not in use this computer was stored in a locked cabinet in the researcher's office. Paper questionnaires were stored in the same manner. Access to computerised data and paper records was available only to the researcher and her academic supervisors.

Parents' interviews were recorded on an iPod. On completion of each interview the recording was uploaded onto the computer that contained the survey data and secured in the same manner. Audio recordings were then deleted from the iPod but remained as voice files on the computer hard-drive. As with the survey data this was available only to the researcher and academic supervisors. Interview transcripts were given the same unique identification code and the parents' survey. All identifying information was removed from interview transcripts including names, locations and named services or institutions.

In the context of the Delphi panel in Phase Two, because this study explored a highly specialised area with a limited numbers of available experts, and used a snowballing sampling strategy, it was possible that participants could logically deduce who their fellow respondents were. In such situations participants cannot remain fully anonymous to each other, although responses should be handled so that their originator cannot be identified, thereby retaining the anonymity characteristics of the Delphi (Vernon 2009). McKenna (1994a) uses the term "quasi-anonymity" when the respondents may be known to each other but their judgments and opinions remain strictly anonymous.

Delphi interviews were treated in the same manner as parents' interviews described above. Delphi questionnaires were assigned a unique identification code so that subsequent round questionnaires with personalised feedback could be provided to individual panel members. All Delphi data were stored on the same computer and secured in the same manner as data returned in Phase One of the study.

In line with the requirements of Dublin City University all data records must be retained for a period of five years following completion of any study. Paper records will be retained in locked storage as described for this period. Voice files will be transferred to CD and stored with paper records. Once this has been performed all voice files will be deleted from the

computer hard-drive. All computer records and CDs will be erased, and paper records shredded, when the five year deadline has expired.

4.8 Conclusion

This chapter has discussed the overarching partially mixed methods design of the current study. The rationale for the choice of a mixed methods approach has been provided, and the design has been related to the aims of each individual Phase, and to the research questions that the study aims to address.

While the individual phases are discussed in greater detail in Chapters Five to Seven the ethical principles on which the study as a whole is based, and the application of these principles to each phase of the study, have been discussed.

Chapter 5: Phase One, Stage One: Parents' Survey & Findings

“Aw, people can come up with statistics to prove anything, Kent,
Forty percent of people know that [*sic*]”.

Homer Simpson quote from episode “*Homer the Vigilante*”
Written by John Swartzwelder, Directed by Jim Reardon
Original air date in North America 6th. January 1994.

5.1 Introduction

The overall mixed methods design of this study and the rationale for its choice has been discussed in the previous chapter. This chapter presents a review of the specific methodology and findings from Stage One of the first phase of the study, namely the parents' survey. The first part of the chapter provides a brief discussion of survey methodology and applies the essential features of the method to the current study. The standardised psychometric instruments for data collection are presented and discussed. These are referred to initially by their full name and thereafter, in the interests of brevity and clarity, referred to in their abbreviated form. The application of the study's theoretical basis to the variables explored in the survey and the instruments used to collect data is described, and the pilot study is discussed.

The second part of the chapter continues with a description of the manner in which the survey data were managed. The statistical tests and text analysis procedures that were performed, and the rationale for their use, are discussed. The results of the data analysis are presented, with a synopsis of the main findings and some preliminary discussion placing these in the context of other published literature. A more general discussion of the findings from the survey will be presented in Chapter Nine where they are integrated with the findings from Stage Two (parents' interviews) and Phase Two (Delphi) of the study. The limitations of this stage of the study are also discussed.

5.2 Survey Methodology

Survey methods are distinguishable from other research methods in terms of the form of data collection and methods of analysis adopted (de Vaus, 2002), although they are not necessarily distinguished by the techniques of data collection which may also be used in other research designs (Calnan, 2007). This study uses a cross-sectional, correlational design which is described by Lavrakas (2008) as involving the collection of data from a sample of individuals at a single point in time, to determine the degree of the relationship between variables for the possibility of making predications based upon these relationships.

5.2.1 Participants and Sampling

In survey designs the samples are not meaningful in and of themselves, their importance lies in the accuracy with which they represent the target population, a good sample being an accurate and efficiently assembled model of the population (Fink, 1995). The critical issues to be addressed in designing a survey sample are those of representativeness and generalisability (de Vaus, 2002; Bruce *et al*, 2008). The target population for this study was parents of children under six with life-limiting neurodevelopmental disabilities. The use of specific diagnostic categories to identify participants for the study was rejected on the grounds that the number of children in any particular diagnostic group would be too small, and such a classification prevented the inclusion of any child who had yet to receive a formal diagnosis (which is possible in the context of very young children). In addition the non-categorical approach used in the study was based on the tenet that the similarities amongst the population as a whole are greater than the specific variations of individual conditions.

The identification of potential participants for the study was complex. The lack of a national database made it impossible to accurately assess the size of the population, which consequently made it impossible to accurately calculate a sampling ratio. Accessing eligible participants was further complicated by the fact that the children were dispersed through a variety of national services and agencies. Initially the NIDD was considered as a potential sampling frame, but the database did not record level of medical need and therefore it was not possible to identify potential participants from this source. The Neurology departments of two major referral children's hospitals indicated that they did not keep a database from which a sample could be drawn for the study, and while they could provide figures for the number

of children attending the departments on an annual basis, it was not possible to identify any demographic, diagnostic or service-related information³. It was also suggested that it would be impossible to identify children who met the inclusion criteria for the study except by manually searching the charts of all children attending the department and, in the context of data protection, it would be extremely complex and time consuming to get approval for this to be undertaken.

Early Intervention Services [EIS] were also considered as a potential sampling frame for the study. The EIS provide a specialist intervention service to young children 0-5 and their families who have been identified as being at risk for developmental delay, and are a single entry point for HSE and voluntary service providers. Unfortunately EIS services are not uniformly available in all geographical areas, entry criteria for services are variable (for example some EIS only take on children who are at least three years old) and many EIS contacted suggested that team leaders would be unable, or unwilling to identify a child as “life-limited”⁴

Although probability sampling would provide a stronger statistical basis for claiming a representative sample, it was not feasible in this study. It is generally accepted that the practice of sampling in social surveys remains problematic in a practical sense. Moser and Kalton (1985:41) suggest that in survey methods “the sample design is decided upon in the light of what is practically feasible as well as what is theoretically desirable”. The realities of social research mean that the ideal samples are often unachievable or unaffordable with many samples having to make do with a range of compromises (Fink, 2003; de Vaus, 2002a; Lavrakas, 2008).

Given the above difficulties this study used purposeful sampling to identify and recruit potential participants. The main objective of a purposeful sample is to produce a sample that can be logically assumed to be representative of the population (Lavrakas, 2008). Participants for the study were recruited through a charity service provider – the Jack and Jill Children’s Foundation. The Jack and Jill Children's Foundation is a registered Irish children’s charity which provides early intervention home respite to families of children with severe

³ Personal communication June 2008

⁴ Personal communication August 2008

neurodevelopmental delay and palliative conditions requiring extensive medical and nursing care, up to the age of five. The Foundation provides national coverage of their services and has a mean annual referral rate of 150 families which they estimate represents 80 - 85% of all children born annually with life-limiting neurodevelopmental conditions. Informal discussions with a significant statutory service provider to the target population corroborated this estimate suggesting, based upon the number of children who are referred to their service from the Jack and Jill Children's Foundation, that the estimate is broadly accurate.

5.2.1.1 Inclusion Criteria

The expert opinion of health professionals experienced in working with the target population was sought in relation to developing inclusion criteria which would indicate a life-limited neurodevelopmental disability. As a result of these consultations families were eligible to participate in the study if they met the following criteria –

- Spoke fluent English.
- Were caring for a child, under the age of six, at home.
- The child had a developmental delay. This may be evidenced by either a diagnosed neurodevelopmental disability or failure to meet age appropriate developmental milestones.
- The child had a medical diagnosis, or complex medical needs, suggestive of a limited life expectancy. These medical needs may include -
 - Children who are sustained by dependence on medical intervention and related technology (for example, tracheostomy / assisted ventilation / enteral or parenteral feeding / administration of intravenous drugs /O2 dependency).
 - Or children who are likely to have any of the following problems over the next year of their life: recurrent pneumonia or other recurrent severe infection; seizure activity that is difficult to control; pain that is difficult to control.

5.2.2 Sampling Procedure

The Jack and Jill Children's Foundation supports up to 250 children and families throughout Ireland at any given time. Analysis of the Foundation's database indicated there were 123

children and families who met the criteria for inclusion in the study. This suggested that the total national population of children and families who would potentially have been eligible for inclusion in the study if a comprehensive sampling frame existed would be in the region of 154⁵. This constitutes a relatively small population. Neuman (2006) suggests that for small populations, defined as less than 1000, the researcher needs a large sampling ratio of at least 30%. Consequently all children and families who met the inclusion criteria were asked to participate in the study. Assuming a response rate of 30% for completed questionnaires suggested that 46 questionnaires would be included in the final analysis. This assumption was based on the response rates reported in studies reviewed in the literature review (Curran *et al*, 2001; Olsson & Hwang, 2006; Thurston *et al*, 2011).

In view of the sensitivity of the subject and the confidential nature of the database, names of potential participants were not passed to the research team. Instead the Foundation forwarded an introductory letter and information booklets to potential participants on behalf of the researcher [Appendices B & C]. The independence of the study from the Foundation was emphasised in the introductory letter. Families were asked to return a completed consent form to the researcher if they agreed to take part in the study. The consent form offered parents the option of completing a postal questionnaire or completing the questionnaire over the phone with the researcher if preferred [Appendix D].

Parents who returned a completed consent form received either a self-administered postal questionnaire or completed the questionnaire over the telephone depending upon their indicated preference [Appendix E]. All questionnaires contained a unique identification code that could be linked to the consent form so that participants were identifiable to the researcher. This was necessary for two reasons; firstly it allowed for survey and interview data to be linked in the final stage of data analysis, secondly it allowed participants demonstrating high levels of psychological distress to be identified and contacted by the researcher so that appropriate action could be taken if required. Completed questionnaires were returned directly to the researcher in a pre-paid envelope.

⁵ Based upon the Foundations corroborated estimate that their database represents approx. 80% of eligible children and families.

In order to maximise response rates to the survey many of the principles of Dillman's "Tailored Survey Design Method" were applied to the study (Dillman, 2002: Dillman *et al*, 2008). However some elements of the design were not implemented, for example the questionnaire did not accompany the information booklet and consent form posted in the first mailing. This decision was taken in the context that the subject matter was relatively sensitive, and it was considered to be intrusive to send the questionnaire without preparation of participants. In addition, it was hoped that adequate preparation, and the salience of the topic, would improve response rates. Only one reminder letter was sent to participants, which was posted four weeks after the original survey. If participants did not then return a completed questionnaire no further correspondence was sent.

5.2.3 Data Collection

Social survey measurement is not error free. Fowler (2009) suggests that the procedures used to conduct a survey will have a major effect on the likelihood that the resulting data will accurately describe what they are intended to describe. Similarly the validity and reliability of instruments used for the collection of data will have a profound effect upon the survey itself (Bruce *et al*, 2008). Bourque and Fielder (2003) recommend that whenever possible questionnaires be either adapted or adopted from other studies. They suggest that there are multiple advantages to such a strategy, particularly in relation to self-administered mail questionnaires, including: the instruments and scales have been developed and tested; the selection of possible answer categories has already been established and tested; instructions have been developed and tested, and they allow data collected to be compared with the findings of prior studies. All of the standardised psychometric instruments used for the collection of data in this study have been widely used and extensively tested in clinical practice.

Quantitative data were collected using a variety of questionnaires. These included: (1) a researcher-developed instrument designed to gather demographic data, and data about current level of need and perceptions and experiences of current services; (2) the Measures of Processes of Care- 20 (King, Rosenbaum & King, 1996); (3) the General Health Questionnaire- 28 (Goldberg & Hillier, 1979); (4) the Multidimensional Scale of Perceived Social Support (Zimet *et al*, 1988); (5) the Impact of Illness on Family Scale (Stein &

Reissman 1980): and (6) the Coping Health Inventory for Parents (McCubbin *et al*, 1983). These instruments were chosen in association with the theoretical framework underpinning the study which was discussed in Chapter One. In this context both the Researcher-Designed Questionnaire and the Impact of Illness on Family scale provide information on the non-normative, cumulative, internal and external stressors experienced by the family. The Measures of Processes of Care- 20, General Health Questionnaire- 28, and Multi-dimensional Scale of Perceived Social Support measure tangible resources which may help ameliorate family stressors, while the Coping Health Inventory for Parents provides information on both effective and ineffective family functioning and adjustment to the demands of the child's condition. The relationship of the constructs measured in the study to the study's theoretical basis is included in Appendix N.

5.2.2.1 Researcher-Designed Questionnaire.

The researcher designed questionnaire was developed to provide the data required to set the context for the study. This included an exploration of the current medical needs of the child, the services that the child and family have available to them, and parent's perceptions of how current services function or might be improved. Items included in the questionnaire arose from reviewing the literature and consultation with clinicians in the relevant field. As this was a new instrument, addressing several distinct aspects of care, expert advice and opinion on its development was also sought. Expert nurses from the fields of intellectual disability nursing, paediatric neurology, and homecare nurses specialising in the care of children with neurodevelopmental disabilities and complex medical needs were consulted. In addition to assisting with the development of the questionnaire this panel of experts critiqued initial drafts and reviewed the final document for content validity.

The questionnaire consisted of 21 questions and was divided into two sections. Section One consisted of eight questions, and collected demographic data and data related to the morbidity experienced by the young child with a life-limiting neurodevelopmental disability. This data related specifically to the difficulties and symptoms experienced by the child, and the parent's perception of how well these difficulties and symptoms were controlled or managed. Questions in this section of the questionnaire were closed or multiple-choice, with space provided for parents to include additional textual commentary if they so required.

Section two of the questionnaire consisted of 13 questions and focused on the services available to children and their families, and on parents' experiences of engagement with these services. This section also included closed and multiple choice questions and again additional space was provided for parents to include any supplemental information they felt was relevant or important.

5.2.2.2 Measure of Processes of Care-20 [MPOC-20]

The Measure of Processes of Care [MPOC] is a measure of family-centered behaviours of health care providers (King, Rosenbaum & King, 1996). It is a self administered, parent completed questionnaire. The instrument was originally developed to examine the way in which care is delivered, and the impact that this service delivery has on children with disabilities and their families. The original MPOC was a 56-item inventory having five factor analytically determined scales; Enabling and Partnership; Providing General Information; Providing Specific Information about the Child; Coordinated and Comprehensive Care for the Child and the Family; and Respectful and Supportive Care.

More recently the MPOC has been refined to a shorter and improved version – the MPOC-20 (King, King & Rosenbaum, 2004). This version retains the same conceptual structure of the five scales that reflect the essential features of family-centred services, but the number of items was reduced and changes made to the format and clarity of response options. The new shorter scale was validated using both the original MPOC-56 data set (653 parents) and a new independent sample of 494 parents. The psychometric properties of the MPOC-20 were shown to be as good as the original instrument using the existing data sets. Cronbach's coefficient alpha was obtained for each of the 5 subscales (range .77 - .88), and test-retest reliability analysis yielded interclass correlation coefficients ranging from .81 - .86.

Concurrent validity was assessed by correlating the MPOC-20 with a Client Satisfaction Questionnaire ($r = .36 - .59$) and with a single item stress variable ($r = -.33 - -.49$). On the independent sample of parents the Cronbach's alpha ranged from .83 - .90. Intercorrelations among the five subscales ranged from .56 - .87 and there were statistically significant correlations between the MPOC-20 and scores on the Client Satisfaction Questionnaire [CSQ] and the Measure of Beliefs about Participation in Family-Centred Services [MBP-FCS] scales.

The MPOC-20 rates responses on a 7 point Likert scale ranging from 1 – *not at all*, to 7 – *to a very great extent*. A scale score is obtained by computing the mean of the rating of the items within each individual scale. Subsequently each respondent yields five scores, one for each of the instruments five subscales. There is no total scale score as the authors considered it more clinically informative to examine the relationship of individual scales to other variables (King, Rosenbaum & King, 1997).

5.2.2.3 General Health Questionnaire-28 [GHQ-28]

The General Health Questionnaire [GHQ] (Goldberg & Hillier, 1979) is a widely used self-administered screening instrument for the detection of a range of psychological disorders mainly in the anxiety / depression spectrum. It concerns itself with two major classes of phenomena: inability to carry out one's normal functions, and the appearance of new phenomena of a distressing nature. The original GHQ was a 60-item inventory although more recently several scaled versions of the instrument have been developed and validated. This study uses Goldberg & Hillier's (1979) 28-item scaled version of the GHQ [GHQ-28].

The GHQ-28 is a self-report measure developed on the basis of the results of principal components analysis of the GHQ-60. It consists of four subscales: somatic symptoms, anxiety and insomnia, social dysfunction and depression (Goldberg & Hillier, 1979; Goldberg & Williams, 2006). Each item consists of asking whether the respondent has recently experienced a particular symptom or item of behaviour on a scale ranging from *less than usual* to *more than usual*. Confirmatory factor analysis confirmed the subscale groupings. Construct validity of the instrument was assessed by correlating the subscales and the total score with Clinical Interview Schedule (somatic symptoms, $r = .32$; anxiety & insomnia, $r = .67$; depression & despondency, $r = .73$). It was not possible to examine the validity of the social dysfunction subscale since ratings of social dysfunction were not made in the interviews. In addition to Goldberg and Hillier's report (1979) several subsequent studies have analysed the validity and reliability of the GHQ-28. Most recently the scale has been investigated as part of the WHO study of psychological disorders in general health care (Goldberg *et al* 1997, Werneke *et al* 2000) and found to be reliable, valid and robust. The GHQ-28 has been extensively tested in practice in diverse populations.

Goldberg and Williams (2006) propose several ways of scoring the scale including simple Likert scoring whereby a weight of 0,1,2,3, is assigned to each column; a “modified” Likert scoring whereby the items are scored 0,0,1,2, a GHQ Scoring System involving weighting the column 0,0,1,1, indicating the presence or absence of the symptom / behaviour, and a discriminant function analysis – a method of scoring which assigns each item a weight directly in proportion to its discriminatory power. An analysis of results obtained by the different scoring methods indicated the GHQ and simple Likert methods to be the most efficient and equally effective. This study used the simple Likert method as the GHQ method considers only the number of symptoms and therefore represents an “area” measure, while the simple Likert method is a composite measure encompassing both area and intensity. In addition this was the scoring method originally used in the validation of the instrument (Goldberg & Hillier 1979), and has been demonstrated to be equally as effective as the GHQ-method by Goldberg *et al* (1997). A total scale score of 23/24 was identified as the best threshold, in terms of the specificity (78.5) and sensitivity (79.8), to determine cases using the simple Likert scoring method (Goldberg *et al*, 1997).

The 28-item version of the GHQ8 was chosen for this study as it has proven to be a psychometrically sound instrument that has been widely tested in clinical practice. It has proven to be less time consuming and simpler to use but equally as effective as longer versions and more complex methods. In addition, it provides more information than other versions of the GHQ which give a single score.

5.2.2.4 Multidimensional Scale of Perceived Social Support [MSPSS]

The Multidimensional Scale of Perceived Social Support (Zimet *et al*, 1988) is a self-report measure of subjectively assessed social support. The scale was designed to measure the perceived adequacy of social support from three sources: family, friends and a significant other. It has been extensively tested in research settings with a variety of population groups.

Zimet *et al* (1988) originally proved the scale to be a valid and reliable measure using a sample of 275 university undergraduates. Confirmatory factor analysis of the scale confirmed the 3 subscale groupings. Cronbach’s coefficient alpha was obtained for each of the 3 subscales (Significant Other $\alpha = .91$, Family $\alpha = .87$ and Friends $\alpha = .85$) and for the scale as

a whole ($\alpha = .88$) indicating good internal consistency, and test-retest values ranged from .72 - .85 indicating good stability. Construct validity of the MSPSS was assessed by correlating the subscales with the Depression and Anxiety subscales of the Hopkins Symptom Checklist ($r = -.25$). Additional support for the MSPSS as a valid and reliable measure was later demonstrated in more diverse populations (Zimet *et al*, 1990; Cecil *et al*, 1995; Stanely, Beck & Zebb, 1998; Clara *et al*, 2001; Bruwer *et al*, 2008).

The MSPSS consists of 12 items which are rated on a seven point Likert scale ranging from 1 *very strongly disagree* to 7 *very strongly agree*. The instrument is scored by summing the individual item scores for the total and three subscale scores, and dividing by the number of items. Each respondent yields four scores, one for each of the subscales and a total scale score. Higher scores reflect higher perceived social support (Zimet *et al* 1988).

Social support has been examined from a variety of perspectives and there are a multitude of social support measurement scales available. However the MSPSS was chosen for this study as it has proven to be a self-explanatory, simple to use, time conserving and psychometrically sound instrument. These were important features in the context of this study, where a number of other instruments were being administered simultaneously.

5.2.2.5 Impact of Illness on Family Scale [IFS].

The Impact of Illness on Family Scale [IFS] was developed as part of a longitudinal study of chronic illness in childhood in an attempt to quantify the impact of this complex domain on the family unit (Stein & Reissman, 1980). The questionnaire was designed to be used as either as a self-report measure or as an interviewer administered instrument.

The IFS has undergone several refinements since its development by Stein and Reissman (1980). Stein and Reissman's original version of the IFS was a 24-item scale yielding a total score measuring total negative impact and four subscale scores measuring financial impact, family / social impact, personal strain and mastery. Internal consistency reliability of the scale and subscales yielded an Alpha Coefficient of .88 for the scale as a whole, and a range of .60 - .86 for the four subscales (Stein & Reissman, 1980). An additional six items measuring impact on siblings were deleted from the original analysis because of missing data

on 50% of the respondents who either did not have other children or had only infants, making the sibling items inapplicable. Subsequent analysis of the scale using data provided from other research projects provided substantial evidence for the generalisability of the instrument.

Following subsequent analysis of the use of the instrument in practice, Stein and Jessop (1985) revised the original scale to a 27-item scale with an additional 6-item sibling subscale. The 27-item scale yields a total score and four sub scores that measure impact on various components of family life. These subscales include: Economic Burden, Social / Familial Impact, Personal Strain, and Mastery, a positive sense of mastery which may emerge from coping with the stress. Internal consistency was high for the scale overall ($\alpha = .83 - .89$), and subsequent analysis also documented that the six items on the original scale that asked about siblings did in fact form a separate subscale ($\alpha = .72$) (Stein & Jessop, 2003). The 1985 review again presented both a one-factor and four-factor solution. The one-factor solution represents a total impact score ($\alpha .88$) which contains the 19 items relevant to the original IFS with the exception of the positively worded mastery items. The four-factor solution yields scores representing general negative impact (10 items, $\alpha .83$); disruption of social relationships (9 items, $\alpha .82$), financial impact (3 items, $\alpha .59$) and coping (4 items, $\alpha .63$).

More recently, following further revision and review of all preliminary data published using the IFS Stein and Jessop (2003) revisited the 27-item scale to further investigate its psychometric properties. Based upon the independent analysis of three large data sets using more diverse samples they propose a one factor, 15-item scale representing general negative impact on the social and family system ($\alpha .83 - .89$) and a second weaker factor-based set of items for financial impact ($\alpha .68 - .79$) and coping / mastery ($\alpha .46 - .52$). Stein and Jessop (2003) advocate that the original scale be replaced by this one factor 15 item total impact factor, and that the revised financial subscale and original sibling subscale be retained as supplementary measures when necessary. This revised scale structure was supported by Williams *et al* (2006) who performed an independent factor analysis of the IFS and found that the psychometric properties of this revised scale were sound when used with a different illness population from that of Stein and Jessop (2003).

This study administered the 27-item IFS in order that all variations of the instrument could be checked for their reliability in the study sample and the most reliable version of the instrument used for the analysis of data. Respondents rank each item on a four point Likert scale ranging from 1 - *strongly agree* to 4 - *strongly disagree*. The total score along with the three subscales – financial impact, social impact and personal strain – measures the negative effect of the child's illness. Negatively worded items are reversed (i.e. items for these three factors are recoded in the opposite direction so that higher scores signaled higher impact). The exception is the coping / mastery subscale in which higher scores indicate greater perceived coping (Stein & Jessop, 2003).

5.2.2.6 Coping Health Inventory for Parents [CHIP].

The Coping Health Inventory for Parents [CHIP] is a 45-item instrument designed to measure parents' responses to the management of family life when they have a child who is seriously and / or chronically ill (McCubbin *et al*, 1983). Forty five coping behaviours are listed and parents are asked to rate how helpful the coping items were to them in managing the home-illness situation. The scale yields three subscale scores which indicate specific coping patterns. These include: maintaining family integration, cooperation and an optimistic definition of the situation (19 items); maintaining social support, self-esteem and psychological stability (18 items); and understanding the medical situation through communication with other parents and consultation with medical staff (eight items).

The instrument demonstrated good internal consistency with alpha values of .79, .79, and .71 for the respective subscales, and the subscales correlated significantly with the Moos Family Environment subscales indicating criterion validity (McCubbin *et al*, 1983). Each item is ranked using a four point Likert scale ranging from 0 - *not helpful* to 3 - *extremely helpful*. Subscale scores are obtained by obtaining the mean of the total of the scores for each item in the subscale. There is no total scale score (McCubbin *et al*, 1983; 1996).

The CHIP has been widely used in research studies related to both acute and chronic childhood illness and with families of children with developmental disabilities which provide further evidence for the reliability of the instrument (Failla & Corson Jones, 1991; Ray & Ritchie, 1993; Woods, Himle & Osmon, 2005; Hobdell *et al*, 2007; Hsieh *et al*, 2009).

5.2.3 Pilot Study

Pilot testing is one of the most critical aspects of a successful survey operation as it provides a safeguard against the possibility that the main survey may be ineffective (Lavrakas, 2008). In any particular survey there may be aspects specific to the population, the subject matter or the data collection methodology that affects the ability of the data collection procedure to perform as intended (Polit & Tatano-Beck, 2009). Moser and Kalton (1985) propose that the most valuable function of a pilot study is to test the adequacy of the questionnaire and the data collection plan.

The questionnaire is one of the critical components in achieving high quality in a survey. Responses may be sensitive to the wording, format and placement of the questions (Kalton & Schuman, 2002; De Maio *et al*, 2002). Foddy (2002) suggests that questionnaires should be tested for comprehension difficulties, perspectives adopted by respondents, and difficulties posed by cognitive tasks in order that revisions or refinements can be made that would eliminate or reduce problems encountered during the pilot test. All of the documentation intended for participants in Phase One of this study were reviewed by the National Adult Literacy Association [NALA] and awarded the Plain English Mark. This ensured they were as accessible as possible for intended users.

There is little consensus amongst researchers with regards to the size of the pilot study. Moser and Kalton (1985:51) propose that “the size and design of the pilot survey is a matter of convenience, time and money”. Five families were chosen to participate in the pilot for the current study. Although this is a small number the decision was made in the context of the small size of the overall population, and the fact that the pilot participants were not being included in the sample which further reduced the number of potential participants for the main study. Families were purposefully selected for participation in the pilot from the sampling frame previously described. They were selected on the basis that they met the inclusion criteria for the study, were able and willing to review and critique the documentation for the study, and were available to be contacted if any clarification of their commentary was required.

During the pilot all documentation for the study was reviewed. This included introductory letters, the information booklet, consent forms, and reminder letters. Participants in the pilot

also completed all data collection instruments, and were asked to provide criticism or commentary regarding the structure and content of the questionnaires. They were also asked to indicate the time taken for completion of the questionnaires and their ease of completion. The goals of the pilot study were to assess: the average time required to complete the questionnaire, the clarity of items, clarity of instructions and the adequacy of the format. There were no major problems identified in the pilot study. Minor revisions were made to the wording and sequencing of some questions but these were not so extensive as to necessitate re-piloting the instruments.

5.3 Data Management and Analysis Procedure

The following section discusses the management and analysis of data. This includes the numerical data from the quantitative instruments, and the textual data from the open-ended questions in the researcher-designed questionnaire and MSPSS.

5.3.1 Management and Analysis of Quantitative Data

Survey data from all of the data collection instruments were coded according to the authors' instructions. Coded data from all questionnaires were entered into SPSS® version 17. Data were screened for errors using descriptive statistics to check the data file for values falling outside possible range for each variable. Following screening of the data, and before any analysis was undertaken, reliability estimates were performed for each scale and subscale. The Cronbach's alpha values computed for each of the data collection instruments in the sample are presented and discussed independently in the Findings section of this chapter. Where indicated both total and subscale scores were calculated for each scale in order that comparisons could be made with the normative values reported by the scales developers, and with scores reported in the published literature.

In order that the most appropriate statistical tests could be performed, data distribution for each scale and subscale was assessed using histograms with a normal distribution curve overlaid to check whether data violated the assumptions of normality. Skewness and kurtosis values and a Kolmogorov-Smirnov statistic were computed, and normal and detrended Q-Q

plots and box plots were examined. Only complete data sets were included in the final data analysis, subsequently the number of respondents reported in analyses differs.

The approach to data analysis depends upon the specific research questions or objectives being examined (Calnan, 2007, Bruce *et al*, 2008). The appropriate choice of a statistical method depends on the extent to which the assumptions about the characteristics and quality of data associated with the method can be met (Fink, 2003). Descriptive statistics including frequency distribution tables, means, and standard deviations were computed to summarise the data. Although the sample was relatively small, and participants were not randomly selected, the data were sufficiently normally distributed to warrant the use of parametric techniques. Streiner and Norman (2008) suggest that ordinal data can be analysed with parametric tests unless it is severely skewed, while Levrakas (2008) suggests that parametric tests are robust to fairly small violations of assumptions of normality. In this context, and relative to the fact that parametric tests were commonly performed in similar studies using these data collection instruments, parametric tests were used to analyse the quantitative data in this survey.

In keeping with the theoretical underpinnings of the study, the total IFS score was considered the dependent / outcome variable. Pearson's product-moment correlations were used for bivariate analysis to explore the strength and direction of relationships between the total IFS score and the study's other continuous variables. As is common in social science surveys the significance level was set at .05 (Streiner & Norman, 2008; Agresti & Finlay, 2009). This level of significance controls the Type 1 error rate however the small sample size does not eliminate the possibility of type II errors occurring (Salkind, 2010; Levrakas 2008).

Independent samples t-tests were used to test for differences in IFS scores depending upon whether the child experienced specific health problems, and whether the family has a named person to coordinate care in the services. Finally One-Way Analysis of Variance (ANOVA) tests were used to test for differences in mean IFS scores between diagnostic groups, the ages of other children in the family and the marital and employment status of respondents.

Multiple regression analyses were performed in an attempt to identify predictor variables for overall negative family impact. Hair *et al* (1995) suggest that in using regression analysis an important decision has to be made regarding the number of predictor variables to include in

the equation. There appears to be no definitive answer to this issue with different authors proposing differing guidelines depending upon the sample size (Stevens, 1996; Tabachnick & Fidell, 2006). While Tabachnick and Fidell (2006:122) suggests that an optimum set of independent variables is the smallest reliable, uncorrelated set that “covers the waterfront” with respect to the dependent variable, Stevens (1996:72) recommends that “for social science research, about 15 subjects per predictor are needed for a reliable equation”, in this context the sample in this study (n=63) allowed for exploration of a maximum of four predictor variables.

There are three major analytic strategies in multiple regression (Hair *et al*, 1995; Tabachnick & Fidell, 2006) with differences dependent upon what happens to overlapping variability due to correlated independent variables, and who determines the order of entry of independent variables into the equation. Standard multiple regression analysis was performed in this study. In this model all independent variables are entered into regression equation at once with each evaluated in terms of what it adds to the prediction of the dependent variable that is different from the predictability afforded by all the other independent variables (Tabachnick & Fidell, 2006). The model was chosen for this study on the basis that the lack of underlying theoretical principles on which to base entry of the independent variables in to the equation out ruled the use of hierarchical regression, and stepwise regression was not suitable on the basis that it can produce a solution that does not generalise beyond the sample unless the sample is large (Tabachnick & Fidell, 2006).

5.3.2 Management and Analysis of Open Ended Questions

Montgomery and Crittenden (2002) suggest that one of the basic measurement problems in survey research is the coding of open-ended questions. According to Lavrakas (2008:140) textual content analysis as it relates to surveys is “a research method that is applied to the verbatim responses given to open ended questions in order to code those answers into a meaningful set of categories that lend themselves to further quantitative statistical analysis”. Green and Thorogood (2009), and Mayring (2000), suggest that the research question and the characteristics of the material should have the priority in the decision about adapted methods for text analysis.

Open ended questions were transcribed into Microsoft Word®. Although these questions were included in the survey in order that participants could, if desired, expand their views or provide additional information, the purpose of analysing this textual data was to merely describe the views of respondents. Consequently the approach to analysis in this study was descriptive rather than interpretative. Additionally, because the textual data obtained from the open ended question were shallow, and supplementary to the quantitative data, in-depth analysis was neither indicated nor possible.

Neuman (2006:323) suggests that there are both qualitative and quantitative versions of text analysis and that in quantitative text analysis “a researcher uses objective and systematic counting and recording procedures to produce a numerical description of the symbolic content in a text”. Textual data in this survey were analysed using this manifest and latent coding framework described by Neuman (2006). In this form of content analysis the answers provided to each open-ended question were explored for the occurrence of recurrent concepts or themes. The themes were counted, recorded and presented numerically as the number or percentage of responses occurring within each theme. Where responses were specific in identifying an issue this was considered a manifest code, for example “appointments can be very inflexible – don’t allow for the fact that you have a very sick child” [I.D.55] was coded “inflexible services”. Where this issue was implied without specifically being mentioned it was considered a latent code and also classified as “inflexible services” for example

“You are given a number of respite hours at the beginning of the year. In the past the person nominated to do the hours was not in a position to do all the hours allocated. There was no one else to do the remaining hours. You are not allowed to carry over the un-used hours to the following year” [I.D.57]

In some instances the textual data supplied by participants were not necessarily relevant to the particular question being answered, but were very relevant to the subject matter of a later question. In these situations the text was transferred to the relevant subject area as described in Figure 5.1. This procedure was performed in order that all data could be included in the final analysis, and to ensure that no information provided by respondents was lost or discounted.

Figure 5.1 *Example of Manifest & Latent Coding of Open-Ended Questions*

Q.19 If you encounter obstacles to obtaining services what effects do these have on family life?"

I.D.	TEXT	CODE ASSIGNED
04	Life can be hard enough without feeling the resentment that my child has not been counted. This affects my personal being and I have been through ups and downs. My other children have worries and are worried about when or if I have to go to hospital as I am their primary carer.	Personal Impact [<i>psychological</i> – life can be hard; resentment; have ups & downs]. Causes worries for other children in family.
13	Everything is a waiting game. You have to find out things for yourself and deal with it. You have to learn things fast, especially medical things that we have no training in. It can be stressful and frustrating when you are not sure what is the right thing to do	Transferred to Q18. Explanation of obstacles encountered. Personal Impact [<i>psychological</i> – stress; frustration; uncertainty]
14	Another hassle. It takes time to make calls, write letters etc. I'm a nurse so I know how the system works but it would be more difficult if you had no background. We have three other small children and we are so busy.	Practical Issues –[burden of time consumed; having to know the system] Family issues – [time taken from other children].

Frequency statistics reported in the analysis of open-ended questions are presented as the number and percentage of times a concept or theme was reported in response to a specific question. As respondents often reported more than one concept, and the number of respondents to individual open-ended questions was variable, the percentages and numbers presented are not consistent with the total number of respondents.

5.4 Survey Findings

The following section reports the results of the data analyses described above. The reliability of the data collection instruments in the sample and the implications for the study is presented at the outset. Descriptive statistics of the sample are then presented followed by a summary of the key patterns that emerged. Inferential statistics are presented followed by a summary of the patterns that emerged from this analysis. Finally the regression analysis is presented and discussed.

5.4.1 Response Rate

One hundred and twenty three families who met the inclusion criteria for the study were identified from the Jack and Jill Children's Foundation database. Five were selected for the

pilot study which resulted in 118 families eligible for inclusion in the main study. All 118 families received a letter inviting participation in the study, together with an information booklet explaining the details of the study and the requirements of participants. Seventy five parents (64%) returned consent forms agreeing to participate.

From these 75 parents, 63 completed questionnaires were returned including five surveys completed by phone with the researcher. This represented 84% of those who agreed to participate in the study and 53% of those originally identified as eligible for participation. Forty five parents also agreed to be contacted by the researcher in relation to participation in a follow-up interview (68%). Telephone participants were not asked to participate in follow up interviews on the basis that they had already contributed a considerable amount of time to the study.

5.4.2 Reliability of the Scales and Subscales in the Sample

The reliability of each scale and subscale was assessed using Cronbach's alpha. Alpha levels ≥ 0.7 were considered "good" according to the criteria set by Streiner and Norman (2008). The reliability of each scale is discussed independently below.

5.4.2.1 Measure of Processes of Care-20

The reliability of the five subscales of the MPOC-20 is presented in Table 5.1 indicating that the scale is reliable in this survey sample.

Table 5.1 *Reliability of MPOC-20 in the Sample*

Measure of Processes of Care (MPOC-20)			
Subscales	Cronbach's α	No. of Items	Valid Responses
Enabling & Partnership	.84 (.63)	3	59
Providing General Information	.87 (.92)	5	60
Child Specific Information	.78 (.79)	3	61
Coordinated & Comprehensive Care	.79 (.81)	4	60
Respectful & Supportive Care	.93 (.90)	5	61
Total Scale	N/A	20	

Values in italics indicate the alpha values reported by King *et al.* 1996.

The alpha values for all subscales in this sample were similar to those originally reported by King *et al* (1996, 1997, 1999) in a sample of children with disabilities, with Siebes *et al*'s (2007) report of the reliability of the subscales in a paediatric rehabilitation sample, and with McConachie and Logan's (2003) assessment of the instrument in a community services context. Although King *et al* (1996) propose that there is no total score for this instrument others have used and reported only the whole scale validity (Sloper *et al*, 2006).

5.4.2.2 General Health Questionnaire-28

The reliability of the total scale and four subscales of the GHQ28 are presented in Table 5.2, and confirm the scale's reliability in this survey sample. Comparison with other studies in similar populations is difficult however as studies have either not reported the reliability of the instrument (Tzoufi *et al*, 2004) or employed alternative versions of the GHQ (Redmond & Richardson, 2003). Lenton *et al* (2001) use the GHQ to explore the parental morbidity associated with childhood life-threatening illnesses but reports neither the version of the instrument used nor its reliability in the study sample.

Table 5.2 *Reliability of GHQ-28 in the Sample*

General Health Questionnaire (GHQ28)			
Subscales	Cronbach's α	No. of Items	Valid Responses
Somatic Symptoms	.84 (.79)	7	62
Anxiety & Insomnia	.87 (.90)	7	61
Social Dysfunction	.83 (.75)	7	62
Depression & Despondency	.92 (.69)	7	61
Total Scale	.93	28	61

Values in italics indicate the alpha values reported by Goldberg & Hillier, 1979

5.4.2.3 Multidimensional Scale of Perceived Social Support

The reliability of the total scale and three subscales of the MSPSS are presented in Table 5.3. The alpha values for both the total scale and subscales indicate that the scale is reliable in the study sample, and compare favourably with the values reported by Zimet *et al* (1988),

Kazarian and McCabe (1991) and Cheng and Chan (2004). The instrument was used by Magill-Evans *et al* (2001) in a sample of families of children with Cerebral Palsy however the study does not report the scales reliability in that sample.

Table 5.3 *Reliability of MSPSS in the Sample*

Multidimensional Scale of Perceived Social Support (MSPSS)			
Subscales	Cronbach's α	No. of Items	Valid Responses
Family	.92 (.87)	4	62
Friends	.93 (.85)	4	61
Significant Other	.96 (.91)	4	62
Total Scale	.92 (.88)	12	61

Values in italics indicate the alpha values reported by Zimet *et al.* 1988

5.4.2.4 Impact of Illness on Family Scale

Due to the various modifications that have been made to the IFS this study administered the 27-item IFS (Stein & Jessop, 1985) so that all possible variations of the instrument could be checked for their reliability in the study sample. From the various editions of the instrument the subscales with the highest reliability in the current sample were used for subsequent data analysis in the study. These subscales are shaded in tables 5.5 to 5.7 which present the results of the reliability of the various forms of the instrument in the sample.

The reliability of the original 24-item IFS and its subscales (Stein & Riessman, 1980) are presented in table 5.4.

Table 5.4 *Reliability of 24- Item IFS in the Sample*

24 Item Impact of Illness on Family (IFS)			
Subscales	Cronbach's α	No. of Items	Valid Responses
Financial Impact	.74 (.72)	4	55
Family / Social Impact	.64 (.86)	9	56
Personal Strain	.69 (.81)	6	61
Coping / Sense of Mastery	.62 (.60)	5	62
Total Scale	.81 (.88)	24	48

Values in italics indicate the alpha values reported by Stein & Riessman, 1980.

Analysis of the Chronbach's alpha for the Family / Social Impact subscale of the 24-item IFS in this sample yielded an alpha of .64 in the sample, considerably lower than the .86 value reported by Stein and Reissman (1980). When the item total statistics for this subscale were examined the reliability of the subscale would have been marginally improved (to .68) by removing the item *I sometimes wonder if my child should be treated "specially" or the same as a normal child*, although this is still lower than the value originally reported for this subscale. A possible explanation for this is that this study excluded incomplete data sets from the analysis whereas Stein and Reissman (1980) and Stein and Jessop (1985) suggest assigning the mean of non-missing items on a particular subscale to missing items on that subscale.

More recent studies using the instrument have used either the 27-item scale, or a modified version thereof. The result of the reliability analysis of the 27-item IFS in this study sample is presented in Table 5.5. However the alpha coefficient for the subscale Disruption of Social Relationships (α .54) in this study is also lower than those subsequently reported in other studies using the 27-item instrument. Examination of the item-total statistics for the subscale indicated that removing any of the individual items resulted in only minimal improvement in the reliability of the subscale.

Table 5.5 *Reliability of 27- Item IFS in the Sample*

27 Item Impact of Illness Family (IFS)			
Subscales	Cronbach's α	No. of Items	Valid Responses
General Negative Impact	.74 (.83)	10	59
Disruption of Social Relationships	.54 (.82)	9	56
Financial Impact	.45 (.59)	4	62
Coping	.62 (.56)	4	62
One Factor Solution (Total Impact)	.80 (.88)	19	48
Impact on Siblings	.95 (.72)	6	57

Values in italics indicate the alpha values reported by Stein & Jessop, 1985.

The Sibling Impact subscale is retained as a separate scale as not all households contain siblings.

Garwick *et al* (2002) reported an alpha value of .87 for the Disruption of Social Relationships subscale of the 27-item IFS in a sample of 99 mothers. However the instrument was modified

to a 17 item version in their study, with the family / social impact subscale reduced to six items. Montagnino and Mauricio (2004) also used the instrument to explore parental stress and coping in a sample of eighteen parents of technology-dependent children with a reported alpha coefficient of .82 for the social relationships subscale. The management of missing data in this study is not discussed.

The low reliability of the financial burden and coping subscales in the current sample reflect the findings of Montagnino and Mauricio (2004). Garwick *et al* (2002) reported an alpha value of .83 for a modified 2-item financial impact subscale.

As Stein and Jessop (2003) more recently advocate that the 27-item scale be replaced with a 15-item scale representing general negative impact on the social and family system, and a second weaker factor-based set of items for financial impact and coping / mastery, the reliability of this latest revision of the instrument was also tested in the study sample. The reliability of this instrument is presented in table 5.6

Table 5.6 *Reliability of Revised 15-Item IFS in the Sample*

Revised Impact of Family (IFS)			
Subscales	Cronbach's α	No. of Items	Valid Responses
Total Impact (15 items)	.78 (.83 - .89)	15	55
Financial Impact	.58 (.68 - .79)	4	55
Coping	.62 (.46 - .52)	5	62
Impact on Siblings	.95 (.72)	6	57

Values in italics indicate the alpha values reported by Stein & Jessop, 2003.

In conclusion, several forms of the IFS have been developed, all of which were tested in this sample before the decision was made regarding which version of the scale to employ in the subsequent data analysis. Based upon the reliability of the various IFS scales and subscales in the sample, the frequency of their use in other similar research projects, and to facilitate comparisons between this and other similar studies, this study uses the 19-item one factor solution proposed by Stein and Jessop (1985) which measures the overall negative impact upon the family, and the financial impact subscale of the 24-item IFS (Stein & Riessman, 1980). The alpha coefficient for this 19-item general negative impact subscale in the current

sample (α .80) compares well with other published reports of the scales reliability in similar samples which range from .81 (Montagnino & Mauricio, 2004) to .88 (Stein & Riessman, 1980; Woods *et al*, 2005).

The original 4-item financial subscale was chosen over revised editions as it had the highest reliability in the current sample (.74 compared to .45 and .58). As recommended by Stein and Jessop (1985, 2003) the sibling items were retained as a separate scale. The coping subscale was not used in the current study due to its low reliability in this and other studies.

5.4.2.5 Coping Health Inventory for Parents

Cronbach's alpha values for subscales I (maintaining family integration, cooperation and an optimistic definition of the situation) and II (maintaining social support, self-esteem and psychological stability) of the CHIP were comparable with those originally reported by McCubbin *et al* (1983) [Table 5.7].

Table 5.7 *Reliability of CHIP in the Sample*

Coping Health Inventory for Parents (CHIP)			
Subscales	Cronbach's α	No. of Items	Valid Responses
Maintaining Family Integration	.78 (.79)	19	56
Maintaining Social Support	.80 (.79)	18	56
Understanding	.68 (.71)	8	57
Total Scale	N/A	24	

Values in italics indicate the alpha values reported by McCubbin *et al*. 1983

An alpha value of .68 was obtained for subscale III (understanding the medical situation) in the current sample. Although this is acceptable it is slightly lower than the value originally reported by McCubbin *et al* (1983) who reported an alpha value of .71 for this subscale.

More recently researchers have tended to modify the scale before reporting the subscale reliability (Katz, 2002; Pei Fan Mu, 2005; Aguilar-Vafaie, 2008). Others have reported alpha values for the scale as a whole rather than for the individual subscales (Ogden Burke *et al*, 1997; Lee *et al*, 2009; Hobdell *et al*, 2007). In some studies the reliability of the scale is not reported (Garro, 2004; Cavallo *et al*, 2009).

5.4.2.6 Implication of Reliability of Data Collection Instruments for the Findings of the Study

Good reliability in the sample was achieved for the MPOC-20, GHQ-28 and MSPSS. Computed Chronbach's alpha values for the subscales, and where indicated the total scale, were at least equal to those reported in the original literature. Although the alpha value computed for CHIP subscale III was slightly lower than the value reported by McCubbin *et al* (1983) this still rounded up to the 0.7 alpha value considered "good" according to the criteria set by Streiner and Norman (2008).

Variable alpha values were computed for the subscales of the IFS depending upon which version of the instrument was used. As previously outlined the subscales with the highest reliability in the current sample were used in the analysis of the survey data. The alpha values for these three subscales all exceeded the Streiner and Norman threshold (2008).

In summary, the analyses of the individual psychometric instruments indicated that they were reliable data collection instruments in the current sample.

5.4.3 Descriptive Statistics for the Sample.

The following section provides a description and summary of the survey data. This includes both the numerical data from the standardised data collection instruments and the textual data from the survey's open-ended questions.

5.4.3.1 Parental Profile and Family Composition

In common with other surveys in similar populations the majority of respondents in this survey were mothers (92%). Respondents ranged in age from 23 to 53 years, and 76% were married. 69% of respondents worked full-time in the home although only 31% of these had given up work specifically to care for their child. An additional 6% had reduced their employment hours to care for their ill child.

The mean number of other children in the families surveyed was 1.4. In 75 % of these families the child with a life-limiting neurodevelopmental disability was either the youngest or only child in the family (n=45). Of these families 52% of respondents reported that they

would consider not having more children because of their child's illness (n=23). Descriptive statistics are displayed in Table 5.8.

Table 5.8 *Parental Profile and Family Composition.*

Variable	Number	Frequency	Valid Responses
Relationship to the Child			63
Mother	58	92.1%	
Father	4	6.3%	
Other	1	1.6%	
Marital Status			62
Single	6	9.7%	
Married	47	75.8%	
Living with Partner	4	6.5%	
Separated	4	6.5%	
Widowed	1	1.5%	
Employment Status			61
Working full-time at home	42	68.9%	
Working full-time outside home	9	14.7%	
Working part-time outside home	10	16.4%	
Other Children in Family			61
Older children only	31	50%	
Younger children only	7	11.3%	
Older and younger children	8	12.9%	
No other children	15	25.8%	

5.4.3.2 Profile of the Children in the Sample.

The mean age of children in the study was three years (range = 1 – 6, S.D.=1.3). Children's diagnostic categories are displayed in Table 5.9. The inclusion of eight children with an unknown diagnosis supports the non-categorical approach to sampling as these children would have been excluded if a categorical approach had been used.

Table 5.9 *Children's Diagnostic Categories.*

Diagnosis	No. of Children	Frequency
Unknown	8	12.7%
Organic CNS Disorder	24	38.1%
Chromosomal Abnormality	24	38.1%
Degenerative Disorder	3	4.8%
Metabolic Disorder	1	1.6%
Combined CNS & Chromosomal	3	4.8%

n = 63

The children experienced a variety of problems which are described in table 5.10. Forty six percent experienced some breathing related difficulty of which 21% were oxygen dependent. Fifty three percent experienced seizure activity. Forty eight percent of the children in the sample experienced some degree of pain or discomfort, in 77% of cases this was due to an identifiable cause. 78% of the children in the sample had difficulties associated with eating and drinking, with 59% of these requiring some form of enteral nutrition. Fifty percent experienced sleeping related difficulties, and in 39% of these cases sleeplessness was associated with a specific symptom (seizures, coughing, apnoea or pain) or a requirement for tube feeding.

Eighty three percent of children experienced communication related difficulties. This included 65% who had little or no verbal communication, and an additional 14% who were profoundly deaf. Almost the entire sample (94%) had mobility associated problems, while 24% had behaviour related problems, and 73% had continence related problems. Additionally 41% of children had other ongoing physical problems which included 46% who had a concurrent chronic physical illness and 42% who were visually impaired. Overall the mean number of problems experienced by children in the study was 5.9 (S.D.=2.2).

Table 5.10 *Profile of Problems Experienced by Children in Sample*

Difficulty Experienced	Number	Valid Percent	Responses
Respiratory Problems	29	46%	63
Unspecified	2	6.9%	
Episodes of Apnoea	2	6.9%	
Oxygen dependent	6	20.7%	
Mechanical assistance required	1	3.4%	
Other respiratory pathology ¹	12	41.4%	
Requires frequent suctioning	3	10.3%	
Aspiration	3	10.3%	
Pain and Discomfort	30	47.6%	63
Unspecified / unknown cause	7	23.3%	
Associated with Gastro Intestinal system ²	14	46.7%	
Associated with Musculoskeletal System ³	9	30%	
Problems with Eating & Drinking	49	77.7%	63
Unspecified	5	10.2%	
Chewing /Swallowing difficulties	7	14.3%	
Requires enteral nutrition	29	59.2%	
Specific / special dietary requirements ⁴	5	10.2%	
Unable to self-feed	3	6.1%	
Sleeping Problems	31	50%	62
Unspecified	4	12.9%	
Excessive wakefulness / requires sedation	15	48.4%	
Related to specific symptom or intervention ⁵	12	38.7%	
Communication Difficulty	52	82.5%	63
Unspecified	6	11.5%	
Little or absent verbal communication	34	65.4%	
Global developmental delay	5	9.6%	
Profoundly deaf	7	13.5%	
Behavioral Problems	15	24.2%	62
Unspecified	6	40%	
Temper or aggressive outbursts	5	33.3%	
Clinging / excessively demanding	1	6.7%	
Autistic tendencies	3	20%	
Mobility Difficulties	59	93.7%	63
Unspecified / Global Developmental Delay	15	25.4%	
Cannot walk	22	37.3%	
Hyper / Hypotonia	13	22%	
Uses wheelchair	4	6.8%	
Gait / Balance problems	5	8.5%	
Continence Difficulties	43	72.9%	59
Unspecified “wears nappies”	40	93%	
Chronic constipation	3	7%	
Other Problems	26	41.3%	62
Visual Impairment	11	42.3%	
Additional chronic physical illness	12	46.2%	
Other	3	11.5%	

NOTE - Percentage figures for each subcategory are based upon the number of positive responses in that category rather than the sample as a whole.

1 refers to all other respiratory conditions including recurrent chest infections / pneumonia / requires frequent nebulisers / asthma.

2 Includes all gastrointestinal system associated causes – abdominal pain / severe reflux / wind / constipation / indigestion.

3 Includes all musculoskeletal system associated causes – muscle pain / cramps / joint pain / movement associated pain.

4 Modification of consistency required – liquidized / pureed / thickened feeds.

5 Symptoms included seizures / respiratory symptoms (coughing / apnoea) / pain / tube feeding.

In addition to specifying the problems their children experienced, parents were asked to indicate how well they felt each of these individual problems were controlled using a simple five point Likert scale (1=very well controlled to 5=very poorly controlled). Generally symptom control did not appear to be problematic. Mean scores for symptom control for each individual problem tended towards the positive end of the spectrum (range from 2.0 to 3.1) indicating relatively good control of individual symptoms [Table 5.11]. This is particularly true in the case of the more acute symptoms amenable to medical intervention for example breathing, seizures, eating & drinking and pain. The more chronic problems, for example sleeping, communication and mobility, tended to have a slightly higher mean score indicating slightly poorer management.

Table 5.11 *Symptom Control in Sample*

Symptom / Difficulty	Mean	S.D.	Min	Max	Valid Responses
Breathing Related Difficulties	2.00	1.02	1	5	28
Seizure Control	2.39	1.31	1	5	31
Pain and Discomfort	2.45	0.95	1	5	29
Eating and Drinking	2.11	1.13	1	5	45
Sleep Related Difficulties	2.90	1.35	1	5	30
Communication Difficulties	3.03	1.24	1	5	47
Mobility Difficulties	3.00.	1.29	1	5	16
Behavioural Difficulties	3.06	1.34	1	5	54
Continence Difficulties	3.00	1.55	1	5	41

Scale range: 1 *very well controlled* to 5 *very poorly controlled*.

An open-ended question was included to provide parents with the opportunity to include additional information about their child's particular difficulties if they wished. Although 39 parents (62%) responded, only 31 responses were relevant to the particular topic of the question. The remaining responses provided information more relevant to the services their child received and the impact this has on the child's difficulties. These responses were re-categorised and analysed in the context of the relevant question.

Of the 31 relevant responses, ten (32%) provided simple straightforward factual information expanding on the problems experienced by the child for example "is suffering from reflux

which causes him to vomit quite often, hence he aspirates and turns into chest infections” [I.D. 18]. However, 48% of responses (n=15) related to the complexity and instability of symptoms and the consequent uncertainty that provokes for parents. Three parents (10%) suggested that problem complexity or problem instability made obtaining outside carers very difficult, while one mother felt that the nature of her child’s illness meant that the child’s care was not seen as a priority by services because “they may come across as a waste of time and effort because there is only so far they can go” [I.D. 04].

5.4.3.3 Service Usage and Perception of Current Services.

The mean number of services involved in the care of children in this study was 8.2 (S.D.=3, range 3 - 16)⁶. In spite of relatively high service usage one in four families (26%, n=15) did not have a named person who they could contact if additional care and support was required and a further 9% of families depended upon a family member to fulfill this role. Where an identified contact person was available to families these came from a variety of sources and services [Table 5.12].

Table 5.12 *Source of Named Contact Person*

Identified Contact Person	Percent	Number	Valid Responses
No One	25.9%	15	58
Social Worker	6.9%	4	
Unspecified Person (service named)	12.1%	7	
Case Manager / Link Person	22.4%	13	
Home Support Worker	15.5%	9	
Community Nurse	3.4%	2	
Paediatrician	5.2%	3	
Family Member	8.6%	5	

Parents’ experiences of services were measured using a simple five point Likert scale [Table 5.13]. Although parents reported that overall they were “fairly satisfied” with the services they received, they also indicated ambivalence regarding several critical aspects of service provision. These included the difficulty experienced obtaining the services needed; the

⁶ This relates to the services of professional disciplines (for example Consultant Neurologist, Physiotherapy, Public Health Nurse) rather than a generic statutory or voluntary service.

frequency with which obstacles were encountered in trying to access services; the level of coordination between services; and the difficulty of accessing information about services. This suggests that while in receipt of services families are fairly satisfied with the service performance, they still experience gaps in relation to service access, information and coordination.

Table 5.13 *Parents Experiences of Current Services*

Variable	Mean	S.D.	Min	Max	Valid Responses
Have difficulty obtaining services needed	3.29	1.29	1	5	62
Have difficulty obtaining information about services	3.27	1.30	1	5	61
How well do services work together	2.86	1.30	1	5	61
Encounter obstacles to getting services	3.06	1.34	1	5	62
Overall satisfaction with services	2.43	1.13	1	5	63

Scale 1 = *very well / very easy* - 5 = *very poorly / very difficult*

Fifty three parents (84%) responded to the request to identify factors that work well in relation to the services they currently receive. Staff attitudes and relationships were paramount in this respect (n = 17). The contribution of charity services was highlighted by thirteen parents, while eleven identified home support services and eight identified the Early Intervention Service as examples of individual services that work well. Other factors that parents perceived as working well included respite services and having an assigned key worker or link person [Table 5.14].

Table 5.14 *Factors That Work Well in Services*

Factor	Percentage of Respondents Reporting Factor	No. of Respondents Reporting Factor
Relationships / Attitudes of Staff	32%	17
Charity Services	25%	13
Home Support Services	21%	11
Early Intervention Service	15%	8
Respite Service	13%	7
Link Person / Key Worker	11%	6
Coordination of Services	9%	5

No. of respondents = 53

Conversely parents also reported the factors that they felt do not work well in current services [Table 5.15]. Fifty respondents (79%) provided information in respect of this issue with the most frequently recurring theme related to service reduction, inconsistency and lack of funding which was reported by almost two thirds of respondents (n = 31). Thirteen parents reported poor coordination of services or poor communication between services, with and an additional five parents reporting difficulty obtaining information about the services available.

Delivery of services was also reported as problematic with parents experiencing long delays and waiting times. It was also reported that a lack of staff continuity impacted upon continuity of care and goal attainment for children, and resulted in the failure to individualise care appropriately. One mother commented -

“I feel at times that the services are aimed for more able-bodied children. Those who really need more and / or are medically challenged are not prioritised. Priority is not a word that should be used but it is necessary for a child like mine” [I.D.04]

The lack of local specialist services and primary care services with expertise in the area of children with life-limiting neurodevelopmental disabilities was also highlighted by five parents.

Table 5.15 *Factors That Do Not Work Well in Services*

Factor	% Reporting Factor	No. Reporting Factor
Service reduction, inconsistency & underfunding	62%	31
Poor communication and coordination of services	26%	13
Long delays & waiting times	20%	10
Lack of individualised care	16%	8
Poor staff continuity	12%	6
Difficult to obtain service related information	10%	5
Lack of local specialist services necessitating long travel periods	10%	5
Bureaucracy	8%	4

n = 50

5.4.3.4 Accessing Current Services

Accessing services was problematic for many respondents in this survey with 50% reporting that it is “fairly” or “very” difficult to obtain the services that they need (n = 31), and a

similar proportion (47%) reported that they encounter obstacles to obtaining services “always” or “most of the time”. Even obtaining information about services was difficult with almost half of respondents (49%) reporting that they found this “fairly” or “very” difficult.

Forty nine parents (78%) indicated a variety of obstacles or challenges they encountered while trying to access necessary services, with eight (16%) using the metaphor of “battle” to describe their encounter with services [Table 5.16]. Principal amongst the obstacles encountered was a perceived overall lack of funding resulting in insufficient or understaffed services. Parents also experienced delays in obtaining necessary equipment, delays with appointments and assessments, and a lack of local area or home based services. Ten percent of parents commented on the bureaucracy they experience in dealing with services particularly in relation to their child’s statutory entitlements. This is exemplified by one parent who reported -

“the Medical Card needs to be applied for annually – this is a waster of time for me and the HSE. [child] needs a medical card – he has a degenerative autoimmune disease and Downs Syndrome. He is not mobile and is PEG fed. He still needs oxygen from time to time and is on 15 different drugs. His injections alone would cost €1300 a day if I had to pay for it. It’s just ridiculous” [I.D. 26]

Table 5.16 *Obstacles Encountered When Trying to Access Services.*

Obstacle	Percentage of Respondents Reporting Obstacle	No. of Respondents Reporting Obstacle
Insufficient or understaffed services	71%	35
Delays in obtaining necessary equipment	20%	10
Appointment / Assessment Delays	14%	7
No local Area / Home Based Services	14%	7
Bureaucracy with Entitlements	10%	5

n=49

Forty five parents (71%) reported on the impact these service related difficulties have on themselves and on family life [Table 5.17]. The predominant theme in this category was the increased personal burden which was reported by thirty two respondents. Although social (life-style limitations and isolation) and physical (exhaustion and ill health) consequences were reported, the major negative impact was psychological. Amongst the negative

psychological effects reported were increased stress and anxiety, frustration and resentment, and anger and depression. At the extreme end of the spectrum this is described by one mother who declared that -

“dealing with services diminishes you – they make you feel so much less than you are. This experience has completely changed me – I feel I’m not the person I used to be” [I.D.40]

Table 5.17 *Impact of Service Obstacles on Family Life*

Impact	% of Respondents Reporting Impact	No. of Respondents Reporting Impact
Personal Impact	71%	32
<i>Psychological</i>	(64%)	29
<i>Physical</i>	(9%)	4
<i>Social</i>	(4%)	2
Negative impact on family unit	56%	18
Direct effect on ill child’s condition	16%	5
Negative effect on relationship with spouse	16%	5
Practical issues / time consuming	16%	5
Financial impact	7%	3

n=45

Other negative effects were also reported. These included a direct effect upon the ill child; a negative effect upon spousal relationships; and the burden of wasting precious time and energy. A negative impact upon the family as a unit was also reported which manifested as insufficient time to spend with other children, strained family relationships, and the experience of a general disruption of everyday family life.

5.4.3.5 Perceived Gaps in Service Provision.

Thirty four parents (54%) identified services they considered would benefit their child but which were currently unavailable to them. In 30% of cases (n=19) this related to the requirement for a greater frequency of services which were already being received, including Physiotherapy (n=11), Speech and Language Therapy (n=11) and Occupational Therapy (n=7). In terms of absent services 6 parents (18%) expressed a need for counselling services for themselves, while 2 parents (6%) expressed the need for counselling services for siblings. An additional ten parents (29%) expressed a need for respite services.

A total of 55 parents identified changes that they believed would improve services for families similar to their own in the future [Table 5.18]. The most commonly proposed change related to improving the availability and delivery of services closely followed by improved frequency and easier access to services. One in four parents proposed the need for easier and improved access to information. In this context seven parents simply suggested that there should be one single source of information on services and entitlements without specifying who or where this should be, but an equal number specified that this should be a designated key-worker who could also assist with coordination of services. Eight parents suggested that families' situations would be improved by the provision of greater home support, while six proposed the need for better respite facilities. Respondents also suggested that greater support is required in the early days following diagnosis of the child's condition. It was suggested that "These are very tough days" [I.D.18] with one parent commenting

"At the time of diagnosis if more information were available and the manner that it was given in and dealt with in a more personal way I feel that the first few years of a child's life and that of their parents would be more bearable" [I.D.31]

Table 5.18 *Factors Considered Necessary to Improve Services to Families*

Factor	% Respondents Reporting Factor	No. of Respondents Reporting Factor
Better Service Provision	44%	24
(Improved frequency / access)	(33%)	18
(Better locally based services & expertise)	(11%)	6
Single source of information/ support	26%	14
Better home support	15%	8
Greater support in early days / diagnosis	13%	7
Respite care	11%	6
Greater humanism from services	7%	4
Better information on rare conditions	7%	4
Home modification	5%	3
Counselling	4%	2

n=55

5.4.3.6 Summary and Interim Discussion of Key Patterns

The profile of families and children in this study is similar to those described in other studies in this area (Lenton *et al*, 2001). Most studies involving children with life-limiting conditions and complex health care needs report that the mother is the main provider of care and more likely to be directly involved in the child's routine medical care (McKeever & Miller, 2004;

Green, 2007a; Monterosso *et al*, 2007; Manaseri, 2008) which possible accounts for the high number of mothers who returned questionnaires.

The children in the sample experienced a wide range of physical problems, and although these incurred significant morbidity parents generally perceived symptom control to be fairly good.

Despite parents reporting that they are fairly satisfied with the services they receive several areas of difficulty relative to services were reported. These included issues of availability, consistence, access and information. Parents also reported negative consequences, both for themselves and for the family as a unit, related to the struggle to access the services they require. There was a high level of service usage amongst the respondents in this study, and a wide variety of services and disciplines were involved in the children's care. Despite this, a significant number of parents did not have a named person that they could call upon to organise additional care and support if required. Overall the service related findings are consistent with those of other studies in this population (Hunt, Elston & Galloway, 2003; Redmond & Richardson, 2003). Both the structure and delivery of services were problematic, with respondents reporting under-resourced and inconsistent services that are difficult to access and insufficient to meet the needs of the population.

The findings from this part of the study accentuate that, in the context of life-limiting neurodevelopmental disability, morbidity is experienced by both the child and the family and services that respond to the needs of both are required. Consequently the need for services to adopt a family focused approach is apparent.

5.4.4 Descriptive Analysis of Standardised Data Collection Instruments

The following section presents a summary and description of the data obtained from the standardised psychometric instruments used for data collection. The skewness and kurtosis of the data are reported to indicate the distribution of the scores amongst the sample for each instrument used. The distribution of scale and subscale scores amongst the sample are compared to the findings the published literature where possible.

5.4.4.1 Measure of Processes of Care

Descriptive statistics for each of the four subscale scores for the MPOC-20 are presented in Table 5.19.

Table 5.19 *Respondents' Scores on MPOC-20 Subscales*

MPOC-20	Mean	S.D	Min	Max	Skew	Kurtosis	No.
Enabling & Partnership	4.62	1.64	1	7	-.41	-.68	59
Providing General Information	3.35	1.61	1	7	.42	-.68	58
Providing Child Specific Information	3.94	1.63	1	7	.14	.31	60
Coordinated & Comprehensive Care	4.70	1.55	1	7	-.45	-.44	59
Respectful & Supportive Care	5.15	1.48	1	7	-.77	-.19	62

Min and Max figures represent respondent's actual scores

Scale Range: 1 = *not at all* to 7 = *to a very large extent*

Respectful and Supportive Care received the highest mean score, while Providing General Information received the lowest. Mean scores for all subscales are slightly lower than the normative scores reported by King *et al* (2004) in a paediatric chronic illness sample, and those reported by Siebes *et al* (2007a) for a paediatric rehabilitation sample. One sample t-test suggested that there was a significant difference between the current sample means and those reported by King *et al* (2004) and Siebes *et al* (2007a) for three of the MPOC subscales including Enabling & Partnership ($p = .03$ & $p = .00$); Providing Child Specific Information ($p = .00$ & $p = .00$); and Coordinated and Comprehensive Care ($p = .01$ & $p = .00$).

There was also a significant difference between the current sample mean score for the subscale Providing General Information and the sample in King *et al*'s (2004) study of pediatric chronic illness ($p = .00$) with respondents in this sample recording a lower mean score. It is possible that the younger age range of children in this sample may be influential in this regard as parents in this study would not have as long a history of service involvement. In addition the rarity of many of the diagnoses in the study sample may influence the availability of child specific information.

5.4.4.2 Multidimensional Scale of Perceived Social Support

Respondents mean scores for the three sources of support measured by the MSPSS and their perceived overall social support are presented in Table 5.20.

Table 5.20 *Respondents' Scores on MSPSS Scale and Subscales*

MSPSS	Mean	S.D	Min	Max	Skew	Kurtosis	No.
Support from Family	5.27	1.55	1.25	7	-.91	.00	63
Support from Significant Other	5.85	1.41	1	7	-1.88	3.84	63
Support from Friends	4.95	1.56	1	7	-.68	.00	62
Total Scale Score	5.38	1.19	1.42	7	-1.13	2.02	62

Scale 1 = *very strongly disagree* to 7 = *very strongly agree*.
Minimum and maximum figures represent respondents' actual scores.

Both subscale and total scale scores in this sample are lower than the normative data reported by Zimet *et al* (1990). The mean score for the total scale in this sample is comparable with the value reported by Magill Evans *et al* (2001) for a families of children with Cerebral Palsy (no individual subscale scores are reported in this study), and similar to the total scale score reported in the same study for a “normal” control group. One sample t-tests indicated that while the difference between the mean scores in this sample was not statistically significant from the score reported by Magill Evans *et al* (2001) for families of adolescents with Cerebral Palsy in their study ($t = -.82, df = 61, p = .42$), there was a significant difference between the mean score for the normal families in Magill Evans *et al*'s (2001) study and the current sample ($t = -.82, df = 61, p = .01$) with the sample in this study reporting a lower mean MSPSS total score. Alternatively the sample mean in this study was significantly higher than that reported by Skok *et al* (2006) for mothers of children with cerebral palsy ($t = 3.08, df = 61, p = .00$)

The MSPPS was the only standardised instrument to offer respondents an opportunity to provide additional information on its topic area. Twenty one respondents (33%) availed of this. Four participants simply expressed how lucky they felt to have good support systems. Three respondents (14%) commented on the social support they received from their

“significant other”. In two cases this was to differentiate between being generally supportive and providing practical support, for example

“My partner is not around very much and rarely participates in my special-need child’s care. Not because he does not want to but he has heavy work commitments. I make all the decisions regarding our special-need child’s care and attend to everything on my own.
[I.D.04]

The third respondent suggested that support from their significant other was lacking due to a conflict in opinion around the treatment options available for their child

The concept of “family support” was qualified by one-third of respondents (n = 7). Four could not avail of family support because they were geographically isolated and had no family nearby, while the remaining three respondents felt that they did not share worries with their families as they did not want to burden them. Similarly four respondents commented that friends “have their own lives” while an additional two respondents expressed that they now have no time for friends. Three respondents commented on the lack of ease or understanding they experience from friends –

“I find my friends are also in their 30’s having their families and are detached from the reality of my everyday life. They are sympathetic but I feel people do remain on a superficial level to what is going on. It’s hard for them to really empathise I suppose – often they don’t even mention [child], it’s like the elephant in the room!” [I.D. 14]

5.4.4.3 General Health Questionnaire

Respondent scores for the total and subscales of the GHQ-28 are presented in Table 5.21.

Table 5.21 *Respondents’ Scores on GHQ-28 and Subscales*

GHQ-28	Mean	S.D	Min	Max	Skew	Kurtosis	No.
Somatic Symptoms	8.1	4.6	0	18	.33	-.76	63
Anxiety & Insomnia	8.4	4.8	0	20	.58	-.11	62
Social Dysfunction	8.0	2.9	4	20	2.04	5.61	63
Depression & Despondency	1.8	3.6	0	20	3.06	11.15	62
Total GHQ-28	26	12.4	8	73	1.2	2.0	62

Scale 0 = *not at all* to 4 = *much more than usual*.
Minimum and maximum figures represent respondents’ actual scores.

Comparison of GHQ-28 subscores with those obtained in other studies is not possible as there has been a tendency to focus on the prevalence of overall psychological morbidity (Lenton *et al*, 2001; Tzoufi *et al*, 2004). Goldberg *et al* (1997) identified the best threshold (in terms of sensitivity and specificity) for identifying psychological morbidity as a total scale score of 23/24 when the Likert scoring method is used. Adopting this threshold (a total scale score of ≥ 24) determines an overall prevalence of psychological morbidity of 46.7% ($n=29$) amongst the sample in this study. However, because the adoption of the cut-off point is central to determination of prevalence, and because previous studies have either not specified the cut-off point (Lenton *et al*, 2001), or employed a different version of the GHQ (Tzoufi *et al*, 2004, Redmond & Richardson, 2003), meaningful comparisons of the prevalence of psychological morbidity between this sample and other studies is impossible.

5.4.4.4 Impact of Illness on Family Scale

The mean scores for each IFS subscale used in the current study are presented in Table 5.22.

Table 5.22 Respondents' Scores on IFS Scale & Subscales

IFS	Mean	S.D	Min	Max	Skew	Kurtosis	No.
One Factor Solution (19-Item Total Impact)	55.96	8.11	37	71	.30	-.56	48
Financial Impact	11.33	3.03	4	16	-.29	-.40	55
Sibling Impact	18.46	7.73	6	24	.56	-1.19	57

Minimum and maximum figures represent respondents' actual scores.

Although the scale is limited by the lack of a designated clinically significant threshold, the mean score is at the high end of the range of potential scores (potential range of scale = 19 – 76). Meaningful comparison with other published reports in similar samples is difficult due to the many variations of the scale used, or the modifications made to the scale by researchers in previous studies (Sheeber & Johnson, 1992; Dodgson *et al*, 2000; Garwick *et al*, 2002; Hsieh *et al*, 2009). The normative data published by Stein & Jessop (1985) used the 24-item version of the IFS scale.

In studies that have used the 19-item version IFS, one sample t-tests indicate that the mean IFS score in the current sample is not significantly different than the score reported by Wright *et al* (2005) in a sample of families of children with Cerebral Palsy ($t = .39$, $df = 47$, $p = .67$). However the sample mean in this study was significantly higher than that reported by Montagnino and Mauricio (2004) for families of children dependent on medical technology ($t = 5.35$, $df = 47$, $p = .00$), and the score reported by Woods *et al* (2005) for families of children with tic disorders ($t = 17.37$, $df = 47$, $p = .00$).

The mean financial impact subscale score is similar to the score reported by Stein & Reissman (1980), but comparisons with later studies are meaningless in the context of the various forms of the subscale used and modifications made to it. Examination of the individual items on the financial impact subscale in this sample demonstrated that 44% of parents ($n = 28$) either agreed or strongly agreed that the child's illness causes financial difficulties for the family.

The mean sibling impact score tends towards the high end of the range of potential scores (potential range of scores 6 – 24). A one sample t-test indicated that the mean sibling impact score in this study is significantly higher than that reported by Stein & Jessop (1985) in their sample of families of children with a range of chronic conditions ($t = 5.23$, $df = 56$, $p = .00$). Sibling impact is not explored in any of the later studies.

5.4.4.5 Coping Health Inventory for Parents

Coping patterns were measured according to the three patterns of the CHIP. The mean scores for each coping pattern are presented in Table 5.23.

Table 5.23 Respondents' Scores on CHIP Subscales

CHIP	Mean	S.D	Min	Max	Skew	Kurtosis	No.
Coping Pattern I (Maintaining Family Integration)	41.48	8.08	20	57	-.19	.04	56
Coping Pattern II (Maintaining Social Support)	30.80	9.89	13	52	-.16	-.96	57
Coping Pattern III (Understanding the Medical Situation)	17.23	3.89	9	24	-.18	-.78	57

Minimum and maximum figures represent respondents' actual scores.

One sample t-tests indicated that the mean score for Coping Pattern I (maintaining family integration) in the study sample was significantly higher than those reported by McCubbin *et al* (1983) for a sample of parents of children with Cystic Fibrosis ($t = 2.47, df = 55, p = .02$); Hobdell *et al* (2007) for a sample of parents of children with Epilepsy ($t = 3.78, df = 55, p = .00$); and Pei Fan Mu (2005) also for a sample of parents of children with Epilepsy ($t = 4.86, df = 55, p = .00$). However, the sample mean was significantly lower than that reported by Lee *et al* (2009) for a sample of parents of children with Asthma ($t = -4.37, df = 55, p = .00$), and Failla and Corson Jones (1991) for a sample of parents of children with developmental disabilities ($t = -3.20, df = 55, p = .00$). There was no significant difference between the mean score in this sample and those reported by Carro (2004) for parents of children with chronic feeding problems, or Cavallo *et al* (2009) for parents of children with physical disabilities.

There was no significant difference between the mean score for Coping Pattern II (maintaining social support) in this sample and the scores reported by McCubbin *et al* (1983), Lee *et al* (2009), Failla and Corson Jones (1991) or Cavallo *et al* (2009). However the sample mean was significantly higher than that reported by Pei Fan Mu (2005) for parents of children with Epilepsy ($t = 6.00, df = 56, p = .00$), and Carro (2004) for parents of children with chronic feeding problems ($t = 4.10, df = 56, p = .00$). It was significantly lower than the score reported by Hobdell *et al* (2007) for parents of children with Epilepsy ($t = -5.19, df = 56, p = .00$).

There was no significant difference between the mean score for Coping Pattern III (understanding the medical situation) in this sample and those reported by Failla and Corson Jones (1991), Carro (2004), and Lee *et al* (2009). The sample mean was significantly higher than those reported by McCubbin *et al* (1983) for parents of children with Cystic Fibrosis ($t = -7.09, df = 56, p = .00$), Pei Fan Mu (2005) for parents of children with Epilepsy ($t = 4.37, df = 56, p = .00$), and Cavallo *et al* (2009) for parents of children with physical disabilities. The sample mean was significantly lower than the score reported by Hobdell *et al* (2007) for parents of children with Epilepsy.

Although Ogden Burke *et al* (1997) also used the CHIP to explore coping patterns in parents of children with chronic & physically disabling conditions the authors report only the mean for the scale as a whole and not subscale means.

5.4.4.6 Summary of Key Patterns from Descriptive Statistics

MPOC-20 subscales demonstrated significant differences in parents' perception of care processes in the current sample compared with previous research in similar populations. The significant differences between the samples mean scores for the four subscales and the published literature indicates less favourable judgments of the care process for each particular subscale in this sample of parents. Providing General Information received the lowest subscale score in the sample with parents reporting that this occurred only "to a small extent". In contrast Respectful and Supportive Care received the highest mean score with parents reporting that this is achieved "to a fairly great extent".

The total GHQ-28 score suggests an overall prevalence of psychological morbidity of 46.7% amongst the sample. Anxiety and Insomnia had the highest subscale score, while Depression and Despondency had the lowest.

Respondents' reported relatively good levels of perceived social support. However the current sample mean was significantly different to Magill Evans *et al* (2001) report of perceived social support in families with "normal" children. Support from a "Significant Other" received the highest subscale score, while support from friends received the lowest.

IFS scores confirm that providing care for a child with a life-limiting neurodevelopmental disability takes a negative toll on the family, and supports the findings of previous research in similar populations. Total negative impact scores were either not significantly different to, or significantly higher than, those reported in previous studies. Siblings also experience a negative impact as a consequence of the family situation, and the mean sibling impact score in this study was significantly higher than that reported by Stein and Jessop (1985). In addition to a general negative impact on the family and siblings, many families experience a negative financial impact associated with the child's condition and ongoing care.

Similar to previous research, the CHIP subscale scores in this sample demonstrate a wide variety of coping strategies employed by parents.

5.4.5 Inferential Statistics

The following section presents the results of the inferential analysis of the data. Only significant correlations are discussed in this section. A full list of all non-significant correlations is included in Appendix O. Correlations are referred to as small ($\leq \pm .29$) medium (.3 - .49) or strong ($\geq \pm .5$) in accordance with the guidelines devised by Cohen (1992), which Field (2009) and Howitt and Cramer (2011) suggests are generally widely accepted. The significance level was set at $p \leq 0.05$.

5.4.5.1 Significant Correlations with Negative Family Impact

Pearson's product-moment correlations were performed to explore possible relationships between overall negative family impact (total IFS score) and the other continuous variables measured in the study including the total and subscale scores for the MPOC-20, GHQ-28, MSPSS and CHIP. Significant correlations are presented in Table 5.24.

Table 5.24 *Notable Correlations with Negative Impact IFS Score.*

Variable	Result (<i>r</i>)	Sig. (<i>p</i>)	N
Factors Related to the Child and their Condition			
Management of sleeping related difficulties	<i>r</i> = -.33	<i>p</i> = .03	47
Service Related Factors			
Frequency with which obstacles to obtaining services are encountered	<i>r</i> = .43	<i>p</i> = .00	47
Satisfaction with services received	<i>r</i> = .39	<i>p</i> = .01	48
MPOC Subscales			
Enabling & Partnership	<i>r</i> = -.35	<i>p</i> = .02	46
Providing General Information	<i>r</i> = -.41	<i>p</i> = .01	47
Providing Child Specific Information	<i>r</i> = -.39	<i>p</i> = .01	47
Coordinated & Comprehensive Care	<i>r</i> = -.33	<i>p</i> = .02	47
Respectful & Supportive Care	<i>r</i> = -.35	<i>p</i> = .02	47
CHIP Subscales			
Maintaining Social Support & Psychological Stability	<i>r</i> = -.31	<i>p</i> = .04	46

The relationship between negative family impact and the management /control of each symptom or difficulty experienced by the child was investigated. The management of sleeping related difficulties was the only difficulty correlated with IFS ($r = .33, p = .03$). Although this was statistically significant, the overall correlation was weak accounting for only 11% of the variance in the data.

Two service related factors were correlated with negative family impact. As might be anticipated there was a moderate positive relationship between negative family impact and the frequency with which parents' encountered obstacles when trying to access services for their child ($r = .43, p = .00$), although the magnitude of the association was weak and it accounted for a modest 18% of the variance. Similarly the positive correlation which existed between negative family impact and parents' satisfaction with the services was also weak ($r = .39, p = .01$) accounting for only 15% of the variance in the data.

Each of the five MPOC-20 subscales was moderately negatively correlated with IFS scores emphasising the importance of the care process in ameliorating the negative impact on families of caring for a child with a life-limiting disability. While the magnitude of this association was strongest for the subscale Providing General Information ($r = -.41, p = .01$) it accounted for only 17% of the variance in the data. The magnitude of the association was even lower for the remaining four subscales with Enabling and Partnership ($r = -.35, p = .02$) accounting for 12% of variance; Providing Child Specific Information ($r = -.39, p = .01$) accounting for 15%, Coordinated and Comprehensive Care ($r = -.33, p = .02$) for 11%; and Respectful and Supportive Care ($r = -.35, p = .02$) accounting for 12% of the variance in the data.

Only one CHIP subscale was correlated with IFS scores. The subscale "Maintaining Social Support and Psychological Stability" (Coping Pattern II) had a weak negative correlation with family impact ($r = -.31, p = .04$). Like the MPOC subscales, the magnitude of this association was small accounting for 10% of the data variance.

Financial impact on the family was significantly correlated with only two variables. This included positive correlations with the frequency with which obstacles were encountered while trying to access services ($r = .34, p = .012$), and with parents satisfaction with the

services they received ($r = .42, p = .00$). The magnitude of these associations was also weak accounting for 12% and 18% of the variance in the data respectively. Although there was a small negative correlation between financial impact and having to give up work to care for the ill child this was not statistically significant.

5.4.5.2 Significant Correlations with GHQ-28 and Subscales

Pearson's product-moment correlations were performed to explore possible relationships between the GHQ-28 and its subscales and the other continuous variables measured in the study. Significant correlations are presented in Table 5.25.

Table 5.25 *Significant Correlations with GHQ-28 and Subscales*

Variable	Result (r)	Sig. (p)	N
Total GHQ-28 Score			
Number of additional services needed	$r = .37$	$p = .00$	62
Somatic Symptoms Subscale			
Number of additional services needed	$r = .45$	$p = .00$	61
Difficulty obtaining information about services	$r = .26$	$p = .05$	61
Level of service integration	$r = .26$	$p = .04$	61
Anxiety & Insomnia Subscale			
Number of additional services needed	$r = .33$	$p = .01$	60
Social Dysfunction Subscale			
Number of additional services needed	$r = .37$	$p = .00$	61
Comprehensive & Coordinated Care	$r = -.32$	$p = .01$	61
Respectful & Supportive Care	$r = -.31$	$p = .02$	62

Total GHQ-28 scores in the sample correlated with only one other variable, namely a moderate positive correlation with the number of additional services required ($r = .37, p = .00$). The magnitude of the association was small accounting for 14% of the variance.

The Somatic symptom subscale was positively correlated with the number of additional services needed ($r = .45, p = .00$), the difficulty experienced in obtaining information about services ($r = .26, p = .05$) and parents perception of the degree of service integration ($r = .26, p = .04$). However the magnitude of these associations was also weak accounting for 20%, 7% and 7% of the variance in the data respectively. The Anxiety and Insomnia subscale also

weakly correlated with the number of additional services needed ($r = .33, p = .01$) accounting for 11% of the variance.

The Social Dysfunction subscale was correlated with three other variables. These included a moderate positive correlation with the number of additional services needed ($r = .34, p = .00$), and moderate negative correlations with two of the MPOC-20 subscales; Comprehensive and Coordinated Care ($r = -.32, p = .01$) and Respectful and supportive Care ($r = -.31, p = .02$). The magnitude of the correlation with the number of additional services needed was strongest, although this accounted for only 12% of the variance. The magnitude of the correlations with the MPOC-20 subscales was even weaker with both subscales accounting for only 10% of the variance in the data.

The depression and Despondency subscale was not correlated with any of the other continuous variables in the study.

5.4.5.3 Additional Significant Correlations

As might be anticipated total MSPSS score was moderately significantly with two CHIP subscales. These included Maintaining Family Integration ($r = .32, p = .02$) and Maintaining Social Support ($r = .30, p = .02$). However the magnitude of the correlation was weak accounting for 10% and 9% of the variance in the data respectively.

There was also a small positive correlation between the total number of problems experienced by children in the sample (which may be taken as an indication of the overall complexity of the child's condition) and the child's age ($r = .28, p = .03$). Although this may possibly suggest that the older the child gets the more problems they experience and by extension the more complex the child's care, this needs to be considered in the context of the small magnitude of the correlation (8% of variance). However, this is supported by the moderate positive correlation between the total number of problems experienced by the child and the number of additional services required ($r = .34, p = .01$), which accounted for 11% of the variance.

The total number of problems experienced by the child was also positively correlated with the frequency with which obstacles to obtaining services were encountered ($r = .27, p = .04$), the overall difficulty in obtaining services ($r = .29, p = .02$) and the level of service coordination ($r = .31, p = .02$). There are two possible explanations for this. The first is that parents experience increasing difficulty as the child becomes older and the care becomes more complex. The second relates to the fact that older children enter the education system and it may be that accessing and coordinating services related to the health and education systems that is difficult for parents. Again these correlations were weak, accounting for 7%, 8%, and 10% of the variance in the data respectively.

No correlation was found between the impact on siblings subscale and any of the other variables in the study.

5.4.5.4 Independent Samples T-tests

An independent samples t-test was computed to compare the mean IFS score between parents who had a named care coordinator available to them and those who did not. A significant difference existed between the mean IFS scores of the two groups ($t = -.27, df = 44, p = .01$). Parents who did not have a named care coordinator available recorded a higher mean negative family impact score ($\bar{x} = 60.4$) than those who did ($\bar{x} = 57.8$). However, using the guidelines proposed by Cohen (1992), the magnitude of the differences between the groups was small (Eta squared = .1).

The availability of a named care coordinator was also used as the grouping variable to explore differences in parents mean scores on service related experiences. These included difficulty obtaining services, difficulty obtaining information about services, how well services work together, the frequency with which obstacles to obtaining services are encountered, and parents overall satisfaction with services. Significant differences in the mean scores between the two groups were found for difficulty obtaining services ($t = -2.5, df = 57, p = .02$) and the frequency with which obstacles to obtaining services were encountered ($t = .82, df = 57, p = .05$). Parents who had a named care coordinator had lower mean scores for difficulty obtaining services ($\bar{x} = 3$) compared with those who did not ($\bar{x} = 4$), and a lower mean score for the frequency with which they encountered obstacles to obtaining

services ($\bar{x} = 2.9$) compared with parents who did not have a named care coordinator ($\bar{x} = 3.7$). The magnitude of this difference was slightly greater for difficulty obtaining services, however, the magnitude of the difference between the groups was very small for both with and effect size .10 and .01 respectively.

Significant results are displayed in table 5.26. Non significant t-tests are included in Appendix P.

Table 5.26 *Results of Independent Samples T-test*

Variable	Result	df	Sig
Mean IFS Score	$t = -.2.7$	44	$p = .01$
Difficulty obtaining services	$t = -.25$	57	$p = .02$
Frequency with which obstacles to obtaining services are encountered	$t = .82$	57	$P = .05$

Grouping variable – Availability of named care coordinator

Respondents were also grouped according to whether the child experienced individual particular medical / physical problems (for example pain) with independent samples t-test used to examine the differences in mean IFS scored between those who experienced a particular problem and those who did not. No significant differences were found between the mean IFS scores in the groups relative to the existence of individual problems with the exception of children experiencing sleeping difficulties ($t = 2.51$, $df = 45$, $p = .016$). Parents whose child did not experience sleep difficulties had a lower mean negative family impact score ($\bar{x} = 53$) compared with parents whose child did ($\bar{x} = 58.7$). The magnitude of this difference was very small (Eta squared = .01).

5.4.6 Regression Analysis: Predictors of Negative Family Impact.

As previously outlined the standard model of multiple regression analysis was used to identify predictor variables for negative family impact. Variables with the highest correlation with overall negative family impact were examined with a view to entry into the regression equation, these included the frequency with which obstacles to obtaining services were encountered ($r = .43$), providing general information ($r = -.41$), providing child specific information ($r = -.39$) satisfaction with services received ($r = .39$), and the availability of a

named care coordinator ($t = -.27$). However because a regression solution is extremely sensitive to the combination of variables that is included in it, and will work best when each independent variable is highly correlated with the dependent variable but uncorrelated with the other independent variables (Tabachnick & Fidell, 2006; Salkind, 2010), the independent variables were first assessed for multicollinearity.

The MPOC subscale “providing general information” was significantly correlated with “providing child specific information” ($r = .53$) with “frequency with which obstacles to obtaining services were encountered” ($r = -.27$) and with “satisfaction with services” ($r = -.34$). Parents’ satisfaction with services was correlated with frequency with which obstacles to obtaining services were encountered ($r = .60$). As Tabachnick & Fidell (2006) identify $r \geq .6$ as the critical level for multicollinearity the independent variable “satisfaction with services” was not included in the regression analysis. Subsequently four independent variables entered the multiple regression analysis, including “frequency with which obstacles to obtaining services were encountered”, “providing general information”, “providing child specific information”, and “availability of a named care coordinator”. Table 5.28 displays the results of the standard regression analysis. Although the model itself proved to be statistically significant ($F = 6.73$, $df = 4$, $p = .00$), it accounted for only a modest 34% of the variance in negative impact on family scores (adjusted $r^2 = .337$).

Table 5.27 *Standard Regression Analysis: Predictors of Negative Family Impact*

Predictor Variable	Standardised Beta	Sig.	95% CI	Part Correlation	% of Variance
Providing General Information	-.17	.26	-.45 – 0.12	-.14	2%
Providing Child Specific Information	-.26	.07	-0.91 – 0.04	-.22	5%
Frequency with which Obstacles to Obtaining Services are Encountered	.25	.06	-0.63 – 3.13	.24	6%
Availability of a Named Care Coordinator.	.32	.02	1.21 – 10.6	.31	10%

When the unique contribution of each of the independent variables was examined only the variable “availability of a named care coordinator” was statistically significant ($p = .02$). However this accounted for only 10% of the variance in negative IFS scores. The remaining independent variables were not statistically significant.

5.4.7 Summary of Key Patterns from Inferential Statistics

In the context of the children in this study and the morbidity they suffer, only one factor significantly correlated with negative family impact. This relates to children experiencing sleep related difficulties, both in terms of the presence or absence of the problem and the degree to which parents perceived the difficulty to be controlled.

Service related factors that correlated with negative family impact included the frequency with which parents encountered obstacles to obtaining the services they needed, their overall satisfaction with services, and all measures of the processes of care. However the magnitude of these associations was weak.

While there were also weak correlations between the GHQ-28 subscales and several aspects of service availability and delivery, only the number of additional services needed correlated with the overall measure of psychological morbidity experienced by the sample (total GHQ-28 score), but again the magnitude of this association was small.

Although the effect size was small, parents who had a named care coordinator recorded lower mean negative family impact scores, less difficulty obtaining services and encountered obstacles less frequently than parents who did not have a named care coordinator.

5.5 Main Findings from Phase One, Stage One

Although quantification of the overall impact on the family of caring for children with life-limiting neurodevelopmental conditions is limited by the lack of a specified clinically significant cut-off point for the scale, the mean IFS score in the sample suggests that care provision has an overall negative family impact. The findings suggest significant emotional and practical stresses experienced by families of children with life-limiting neurodevelopmental disabilities, and text analysis corroborates this finding with several aspects of family impact reported including strained family relationships and a general disruption of everyday family life.

Managing the treatment of a child with a life-limiting neurodevelopmental disability involves a complementary relationship between the healthcare team and the child's family. The survey findings suggest that it is these process related variables, and the nature of parents relationship and engagement with services, that have the most significant impact on families caring for children with life-limiting neurodevelopmental disabilities. While the severity of the child's condition did not significantly correlate with the negative family impact score, the manner in which services are coordinated, their ease of access, and the process by which they interact and engage with parents are all significantly correlated with the family impact. Although lack of services (as indicated by the number of additional services needed) did not directly correlate with family impact it did correlate with parents' psychological morbidity.

Parents in this survey were clearly able to differentiate between the services they actually receive, and their perceived overall requirements. For although parents' reported that they are fairly satisfied overall with the services they do receive and identify aspects of service provision that work well, they also report gaps in both service provision and service delivery. The findings suggest that current services to children with life-limiting neurodevelopmental disabilities and their families are characterised by service reduction, inconsistency and lack of funding. Coordination and communication were identified as problematic and obtaining information about services was identified as an ongoing challenge for parents.

Overall the findings suggest that there are not high levels of unmet physical needs amongst the children in this sample, for although the children experienced a significant number of physical problems, parents generally considered these to be fairly well controlled. This supports the assertion that specialist palliative care services are not required for all children with life-limiting disabilities.

Predicting family impact is difficult and the predictor variables are nebulous. The results of this study highlight the complexity of the experience for parents as they attempt to navigate the healthcare system and access the services they require. The findings are important as many of the variables and issues identified as significant in terms of negative family impact can be manipulated without significant fiscal implications.

5.6 Preliminary Discussion

This section presents a provisional discussion of the overall findings of the quantitative data analysis. The findings will be discussed in greater detail in Chapter Nine where they will be integrated with those from subsequent stages of the study, and with those of previously published research.

The response rate in this study compares generally with the rates reported in the literature for similar surveys with this population which, when reported, range from 29% (Hunt, Elston & Galloway, 2003) to 73% (Lenton *et al*, 2001). Some studies do not report an overall response rate indicating only the number of study participants. Again, the number of participants in this study compares reasonably well with reported figures which range from 17 for a generic neurodevelopmental disability population (Redmond & Richardson, 2003), to 129 for a more diverse comparative study of malignant and non-malignant life limiting childhood illness (Monterosso *et al*, 2007). Based upon the number of children on the Jack and Jill Children's Foundation database, and extrapolation of this to estimate the number of children nationally who would have met the inclusion criteria as previously discussed, this response rate meets Neuman's (2006) criteria for a sampling ratio for small populations. The overwhelming majority of respondent in this study were mothers, and although this may be perceived as a limitation of this stage of the study is unsurprising in the context that often the majority of the practical burden falls on mothers who most frequently take care of the child at home (Mastroyannopoulou *et al*, 1997; McKeever & Miller, 2004; Green, 2007a; Monterosso *et al*, 2007; Manaseri, 2008; Nicholl, 2008).

Morbidity experienced by children in the sample is similar to that reported by Lenton *et al* (2001) although Lenton *et al*'s study did not report on symptom control. The extent and nature of the children's problem indicated that they require highly complex care, and this is consistent with the findings of other studies exploring the care requirements of children with life-limiting conditions and complex disabilities (Roberts & Lawton, 2001; Redmond & Richardson, 2003; Quinn *et al*, 2005; Steele & Davies, 2006; Nicholl, 2008). However overall the findings suggest that there are not high levels of unmet physical needs amongst the children in this sample, for although they experienced a significant number of complex physical problems parents generally considered these to be fairly well controlled, supporting

the position that specialist palliative care is not required for all children in this population (DOH&C, 2010). Symptom management had not been reported in previous studies.

The current sample mean score for perceived social support was significantly different to Magill-Evans *et al* (2001) report of perceived social support in families with “normal” children. Social support from respondents’ “significant other” received the highest mean score; however this should be interpreted in the context of the accompanying textual commentary which suggests that this may not necessarily translate into practical help and support in the day-to-day care of their child. Social support from friends received the lowest mean score in this sample. This is unsurprising in the context that the time-consuming care often required by children with life-limiting neurodevelopmental disabilities may leave families with little time for those outside the immediate and extended family (Alexander *et al*, 2002; Steele & Davies, 2006; Murphy *et al*, 2006; Nicholl, 2008; Manaseri, 2008) In addition respondents’ comments in this study also suggested a reluctance to burden friends who “*all have their own lives*” or who may be ill at ease with the complexity of the child’s condition.

However, the lack of a significant correlation between perceived social support and psychological distress (total GHQ-28 score) or family impact is both surprising and contrary to the findings of other findings. Childhood disability literature identifies social support as an important factor in the stress experienced by families of children with disabilities (Taanila *et al*, 2002; Kelso, French & Fernandez, 2005; Glidden, Billings & Jobe, 2006; Giallo & Gavidia-Payne, 2006; Glidden & Natcher, 2009).

It is possible that this anomaly may be a function of the myriad of ways in which both psychological distress and social support is construed and measured: both Taanila *et al*, (2002) and Kelso, French and Fernandez (2005) use a qualitative approach to describe the construct; both Glidden, Billings and Jobe (2006) and Glidden & Natcher (2009) use a subscale of the Ways of Coping Questionnaire; Giallo and Gavidia-Payne (2006) use a subscale of the Self-Report Coping Scale; and Larson used a combination of interviews and coping scales. It may also be possible that the magnitude of the burden of practical care that families of young children with life-limiting neurodevelopmental endure may mediate the positive effects of social support.

GHQ-28 scores indicated that the overall prevalence of psychological morbidity in the sample is similar to other published reports. Tzoufi *et al* (2005) reported a prevalence of psychiatric morbidity of 43.2% amongst fifty two parents of children with chronic neurological disease, while Lenton *et al* (2001) and Mastroyannopoulous *et al* (1997) reported a prevalence of 53.8% and 54% respectively for mothers of children with life-threatening conditions. Depression and Despondency had the lowest mean subscale score in the sample while Anxiety and Insomnia had the highest mean subscale scores for participants in this study, indicating that psychological morbidity in the current sample tended to manifest as anxiety rather than depression.

Comparison of the rates of psychological morbidity in this sample of mothers with normative data from the general Irish population is difficult due to the variety of assessment tools used; however it would appear that the prevalence is much higher in the current sample. A recent national study by the DOH&C (2009b), using the Mental Health Inventory-5 [MHI-5] as a diagnostic tool reported that 7.5% of adult women were identified as having high levels of psychological distress sufficient to cause a “probable mental health problem”. Using the Composite International Diagnostic Interview Short Form [CIDI-SF] the same study reported Generalise Anxiety Disorder among 3% of the women in the survey. While a recent telephone survey of 2,711 people using the GHQ-12 as a measure of psychological distress conducted by the Health Research Board [HRB] found that 12% of respondents had probable mental health problems (Tedstone Doherty *et al*, 2007).

Despite relatively high service usage over a quarter of the families in this study could not identify a named person to call upon to coordinate services and support. Generally the service related findings of this study generally support those of previous national and international studies in this population (Lenton *et al*, 2001; Kirk & Glendinning, 2002; Hunt, Elston & Galloway, 2003; Redmond & Richardson, 2003; Tzoufi *et al*, 2005; Quinn *et al*, 2005; Monterosso *et al*, 2007). Respondents reported under-resourced and inconsistent services, which are difficult to access, bureaucratic, and insufficient to meet the needs of the population.

Mean scores for MPOC subscales provide additional useful summary statistics about the relative strengths and weaknesses of current services to children with life-limiting

neurodevelopmental disabilities and their families. It would appear that the strongest aspect of current services (as indicated by the highest mean subscale score) is related to the type of interaction and relationship services have with families rather than to any pragmatic aspect of service provision. This is congruent with parents' identification of the factors that they consider work well in the services they receive where relationships as attitudes of staff was the predominant theme. However both subscales scores (Respectful & Supportive Care and Enabling & Partnership) are located around 5, indicating that in general parents report that services meet this need "*to a fairly great extent*" suggesting that there is some room for services to improve in this respect.

The MPOC subscale Providing General Information received the lowest mean score indicating that it is perceived by parents to be the weakest aspect of current services. In general parents reported that their general information needs were met only "*to a small extent*" by current services. This supports the findings of previous studies which suggest that accessing information is a consistent challenge for parents (Lenton *et al*, 2001; Mastroyannopoulous *et al*, 1997; Redmond and Richardson, 2003; Smith Stepanek, 2009). The relatively low mean scores for this subscale and for the subscale related to coordinated & comprehensive care corroborates parents textual comments related to factors that do not work well in the services they receive.

5.7 Strengths and Limitations of Phase One, Stage One

The strengths and limitations of this stage of the study should be considered before conclusions can be drawn. The main strengths of this stage are its clearly defined sample, both in terms of the children that were included and the specific age range that was investigated. In addition this study addresses a population of children and their families that, up to this point, have generally not been explored as a unique and individual group. The main limitations of this survey include the size of the sample and the sampling frame from which it was drawn, and a reliance on self-report measures predominantly from the mother.

This survey used a relatively small sample. The absence of any empirical data about the size of the population made it impossible to perform an accurate power analysis, and the sample

size may have influenced the power of the statistical analysis performed. Despite this, the sample size does meet Neuman's (2006) criteria for sampling small populations.

The study used the database of a national children's charity to recruit participants. Consequently all of the respondents in this survey were, at the time of the study, in receipt of the services of this charity. This included home-nursing hours and home visits from specialist nurses. These families therefore had additional support services beyond those that may be available to the general population of young children with life-limiting neurodevelopmental disabilities and their families. In addition the study depended upon parents volunteering to participate and it is possible that this may have biased the study to better adjusted families. However, the fact that this is a national children's charity meant the survey obtained national coverage rather than the site specific samples used in many previous studies.

All of the families recruited to participate in this stage of the study were identified by service providers which could potentially introduce a bias into the sample. In order to minimise this the inclusion criteria for the study are explicit, and were agreed with expert practitioners as indicators of a limited life expectancy.

5.8 Conclusion

This chapter has provided an overview of the quantitative stage of this mixed methods study. The chapter began with a brief overview of quantitative research and the issues to be considered in utilising a survey approach, and proceeded to place the current study in this context. The research design, its implementation, and issues of quality have been discussed. The method of data analysis has been described.

The findings of this stage are generally consistent with previous national and international research in the area. The findings support the view that young children with life-limiting neurodevelopmental disabilities require care that is complex and skilled. Providing this care has consequences beyond the child and impacts on all aspects of family life. Mothers report many challenges associated with the services available to these children and their families which appear to add to the overall burden of care.

Chapter 6: Phase One, Stage Two: Qualitative Methodology & Findings

“If scientific reasoning were limited to the logical processes of arithmetic, we should not get very far in our understanding of the physical world. One might as well attempt to grasp the game of poker entirely by the use of the mathematics of probability”.

Vannevar Bush (1945)

Cited in: Wardrip-Fruin N, Montfort N. (2003) *The New Media Reader*. MIT Press; Cambridge, MA. p.42

6.1 Introduction

Sarantakos (2005:53) suggests that quantitative research can “result in “meanings” that are closer to the beliefs of the researcher than to those of the respondents”. Bryman (2006) concurs, proposing that one facet of the distinction between quantitative and qualitative research is that the former is orientated to the specific concerns of the researcher and the latter to the perspectives of the research participants. While the survey undertaken in Stage One provided a quantifiable overview of important facets of the provision of care to young children with life-limiting neurodevelopmental disabilities and their families, its objective nature did not permit the opportunity to develop a deeper understanding of parents’ experiences and perspectives. For this a more subjective interpretivist strand was required. In order to provide a comprehensive insight into the issues involved in caring for a young child with a life-limiting neurodevelopmental disability, and to keep participants’ perspectives central to the study, an additional qualitative approach was required.

This chapter presents the methodology and findings from the parents’ interviews which formed the second stage of Phase One of the study. Data analysis and quality issues are discussed, and the findings are presented with some preliminary discussion. Findings are discussed in greater detail where they are integrated with those of the other phases of the study in Chapter Nine.

6.2 Sampling Parents for Interview

Congruent with an explanatory mixed methods design, parents for interview in Stage Two were selected from the respondents to the parents' survey. Although the overarching design for sample selection was purposive, within this framework a modified random selection procedure was employed to select interviewees.

Participants were eligible for inclusion in this stage of the study if they had returned a consent form indicating their agreement to be interviewed with their completed questionnaires and had a total Impact on Family score [IFS] from Stage One. IFS scores were used as the basis for selecting parents as it was used as the outcome variable in Stage One. This resulted in the identification of 41 potential parents for interview. Gillham (2005) suggests that deciding on the number of interviews to conduct depends upon whether the technique is being used as a preliminary exploratory stage, or to develop a framework of explanation. Unlike quantitative research, there is no empirical method for calculating the number of participants required for a qualitative study. As such, the decision was made to initially interview 12 participants representing 30% of those eligible. This number of interviews is generally consistent with the number of parents interviewed in previous qualitative studies exploring parents' experiences of providing care to their child with complex needs (O'Brien, 2001; Steele & Davies, 2006; Green, 2007a). An additional six parents were selected, but held in reserve, to be interviewed if data saturation did not occur with the first 12 interviews.

In order to select parents for interview the range and distribution of IFS scores amongst the sample was inspected. IFS scores were categorized as low impact (31 – 39); middle impact (40 – 48) and high impact (49 – 58) and the proportion of the sample in each category was computed. Twenty percent of parents fell within the low impact category, 51% within the middle category, and 29% within the high impact category. These proportions were used as the basis for interview selection.

The 41 parents were allocated to the low, middle or high impact categories according to their IFS score. From these categories two parents were randomly selected from the low category (20%), six from the middle impact category (50%) and four were selected from the high

impact group (30%) representing the proportionate distribution of IFS scores amongst the parents sample as a whole [Appendix Q]. An additional two parents were selected from each category to be held in reserve in case they were needed for data saturation.

6.3 Data Collection

There are several established qualitative research methodologies and consequently several methods of data collection. Wolcott (1998:10) succinctly categorises three major modes through which qualitative researchers gather their data: participant observation (experiencing); interviewing (enquiring); and studying materials prepared by others (examining). In this study narrative data were collected through interviews which can facilitate rich description and detailed accounts of participants' experiences and perspectives (DiCicco-Bloom & Crabtree, 2006) with interviewing having a strong claim to being the most widely used qualitative research method (Fielding & Thomas, 2008; Green & Thorogood, 2009). There are many approaches to qualitative interviewing which differ in terms of the breadth and focus of the interview (Rubin & Rubin, 2005; Gillham, 2005; Rapley, 2005). Fielding and Thomas (2008) and Gibson and Brown (2009) provide a typology of qualitative interviews based upon the degree of structure or standardisation imposed by the interviewer. This study used semi-structured interviews to collect data. A semi-structured interview is a valuable exploratory tool and involves a set of predetermined but open-ended questions that allow for spontaneous and in-depth responses (Ryan, Coughlan & Cronin, 2009). Gillham (2005) proposes that this form of interview is particularly useful where the person being interviewed may be inhibited or constrained by a more structured approach, or where the interest is in some dimension of the individual's life-experience, and where significant themes can only be elicited by allowing the individual to give their account in their own way. In addition, Fielding and Thomas (2008) suggest that semi or unstructured interviews are especially useful when the research topic is sensitive or complex as is the case in this study. Rubin and Rubin (2005) outline the process of conducting semi-structured research interviews which includes the development of the interview guide, conducting the interview and analysing the interview data. This process will now be discussed in the context of the current study.

6.3.1. Developing and Piloting the Interview Topic Guide

Ryan, Coughlan and Cronin (2009) suggest that developing the interview topic guide is central to obtaining data that will address the study's purpose and objectives. The topic guide used for interviews in this study was informed by several sources including: the study's research questions and overall aims, the literature review, and the data collected from parents in Stage One [Appendix R]. The questions focused upon the three areas central to the study – the child, including the challenges and difficulties experienced; the family, including the impact of care provision on other members and the family unit and the management of day-to-day life; and the family's engagement with and experiences of the services available to them. The guide was composed of broad, guiding questions related to these three areas, and was designed to elicit descriptive responses and stories from participants. These guiding questions were supplemented and supported by prompts and probes when appropriate to encourage participants to expand upon an answer or to redirect them back to the main topic if they became sidetracked and lost focus.

Once the topic guide was developed a pilot study was conducted. The pilot study served two purposes. It allowed the interviewer to assess the comprehensiveness of the topic guide, and facilitated identification of any difficulties with the interview process itself. The parent interviewed in the pilot study was randomly selected from the list of those eligible to participate in the interviews as outlined above. There were no issues identified in the pilot study and subsequently no changes were made to the topic guide or interview process. The pilot interview lasted 50 minutes, providing an indication of the potential duration of subsequent interviews. The decision was made to include the transcript from the pilot interview in the final data analysis on the basis that no changes were made to the topic guide or interview process, the considerable time and effort the mother had committed to the study, and relatively small number of parents eligible for interview overall.

6.3.2 Negotiating the Interview and Preparing the Interviewees

Once identified, the researcher contacted potential interviewees by telephone. From the twelve parents originally selected two were no longer in a position to participate (one parent's child was seriously ill in hospital, the other child had died two months previously). Two additional parents were selected from the reserve list previously described. All parents

who were contacted agreed to be interviewed for the study. Obtaining initial verbal consent over the telephone provided the researcher with the opportunity to reiterate the information provided at the outset of the study. This allowed parents the opportunity to reflect upon this information, and consider any questions or reservations they may have well in advance of the interview taking place. As eight months had passed since the initial contact about the study had been made, parents were reminded of the following issues at this point of telephone contact –

- The purpose of the study.
- The parameters of the interview. Parents were informed of the issues to be discussed and verbally presented with the interview topic guide.
- Parents were informed that it would be necessary to record interviews and the handling of audiotapes and interview transcript was discussed.
- Pragmatic aspects of the process were discussed for example the time required based upon the duration of the pilot study, choice of interview venue, and the need for a relatively distraction-free time period.

Gillham (2005) suggests that preparing interviewees in advance addresses both pragmatic and ethical dimensions. It allows interviewees an opportunity to reflect on whether or not they agree to the topic, and a chance to reflect on what they might say and prepare themselves. An interviewee's clear understanding of what they are being asked to do, and how what they say will be treated, are fundamental in setting the tone of the interview which subsequently influences the confidence and candor of the respondent. At the end of the telephone conversation an appointment was made to conduct the interview at a time and place convenient for the parent.

Ten of the twelve parents requested that they be interviewed in their home; two requested that the interview be conducted over the phone. All parents needed to fit interviews around their child's schedule of appointments and services, and in some instances the interviews were scheduled for over a month later. Parents were given a phone number on which to contact the researcher should they need to cancel or reschedule the interview, or if they had any questions they wanted to discuss in advance of conducting the interview. Five of the twelve interviews needed to be rescheduled due to either a crisis in the child's condition or an

unanticipated hospital appointment. Three of the five were rescheduled more than once. The challenge to initially schedule interviews with parents was representative of the complicated and complex routines that parents who care for young children with life-limiting neurodevelopmental disabilities experience.

All parents were contacted two days before the scheduled interview date to confirm that they were still willing and able to participate.

6.3.3 Preparing for the Interview

To prepare for each individual interview the data returned by the parent in Stage One were thoroughly reviewed. This ensured the researcher was familiar with the data the parent had already provided making certain that they were not asked to repeat information twice. It also allowed the researcher to identify aspects of the survey data on which clarification was required.

6.3.4 Conducting the Interview

Astedt- Kurki and Paavilainen (1999) suggest that clear explanations of what to expect ease the interview process. Before conducting the interview parents were reminded of the topic, format, purpose, and need to record the interview. The importance of parents' individual experiences to the more general picture was emphasised. The procedure for handling audiotapes and interview transcript was reiterated. Finally, parents' questions were addressed, and written consent for participation was obtained.

Although interviewees were given an expected timeframe for the interview of approximately one hour based on the pilot, in reality this was not the case when conducting the remainder of the interviews. The time taken to conduct the interviews ranged from 40 minutes (telephone interview) to a little over three and a half hours depending upon what the interviewee wished to share, and the number of interruptions to the interview. In all but two face-to-face interviews the child was present and interviews were interrupted to allow the mother to attend to the needs of the child. This included starting or finishing PEG feeds, providing suctioning for respiratory difficulties, administering medications, or attempting to soothe an unsettled

child. Frequently the interview proceeded with the mother holding or carrying the child. In the remaining two interviews the child was being cared for by an in-home support worker, although again the interview did not proceed uninterrupted in that the parent frequently needed to field inquiries and difficulties. Interviewing in the home provided a rich opportunity to see first hand the context in which parents provided care, and in many cases the child for whom this care was being provided.

These interviews explored issues of profound importance to participants. In conducting the interviews the researcher was mindful that she was being given, in trust, private and sensitive information about the child and the family, and was constantly aware of the ethical responsibility she had to interviewees in the study. Although a research interview is not a therapeutic interview Donalek (2009) cautions family researchers to remember that they enter a sacred space in which the most intimate, formative and sustaining processes of human existence take place. Gillham (2005) suggests that people are responsive to the apparent interest of interviewers and therein lays the essence of their vulnerability.

The researcher was aware that the process could potentially be distressing for parents. It was possible that interviewees could be distressed by the process or the topics to be discussed during the interview, or alternatively could later regret disclosure. The procedure for dealing with these situations has already been described in Chapter Four. Three parents became upset during the interviews. Where an interview topic appeared to be a cause of difficulty for a parent they were offered the option of discontinuing the interview or taking a break. All parents wanted to continue with the interview. The researcher remained with, and supported, the interviewee after the interview as needed (Astedt- Kurki & Paavilainen, 1999; Horowitz, Ladden & Moriarty, 2002). All ten parents who participated in the face-to-face interviews even those who appeared to have had some difficulty during the interview itself reported that they appreciated the opportunity to discuss the topic openly.

6.4 Management of Interview Data

All interviews were recorded on an iPod with parents' permission. Audio recordings were uploaded onto computer and used to make verbatim transcriptions into Microsoft® word documents. These transcripts formed the basis of data analysis. Interview transcripts were

given the same unique identification code as the parents' surveys returned in Stage One, and were then anonymised with any identifying names of individuals, institutions and locations removed. In view of the relatively small size of the population, and the rarity of some of the children's conditions, children's diagnoses were also removed from the transcripts in order to minimise the potential for parents to be identified. The audio recording was deleted from the iPod once it had been uploaded onto a secure password-protected computer.

6.5 Data Analysis

In qualitative research data analysis may involve a variety of interconnected interpretative practices which vary in their underlying epistemological assumptions (Denzin & Lincoln, 2005; Seale *et al*, 2006; Lyons & Coyle, 2007). Green and Thorogood (2009) propose that the decision about which qualitative data analysis method to use is derived from the needs of the study. Braun and Clarke (2006) suggest that qualitative analytical techniques can be roughly divided into two camps: (1) those that are affiliated to a particular theoretical or epistemological position where there is limited variability in how the analytical method is applied within the framework (for example Conversation Analysis, Interpretative Phenomenological Analysis) including those where there are differing manifestations of the method from within the broad theoretical framework (Discourse Analysis, Grounded Theory); and (2) those which are essentially independent of theory and epistemology and can be applied across a range of qualitative approaches. Qualitative Thematic Analysis is positioned in this second camp.

Thematic analysis is a common general analytical strategy for qualitative data which facilitates the search for patterns of experience within the data set. Given (2008:867) defines thematic analysis as "a data reduction and analysis technique by which qualitative data are segmented, categorised, summarised and reconstructed in a way that captures the important concepts within a data set". Holloway and Todres (2003:347) identify "thematizing meanings" as one of a few shared skills across qualitative analysis. Boyatzis (1998) suggests that rather than a specific method or approach in itself thematic analysis is a tool, or approach, performed within major analytical traditions. However Braun and Clarke (2006) propose that thematic analysis should be considered a method in its own right as its

theoretical freedom provides a flexible tool which can provide a rich and detailed yet complex account of data.

The product of a thematic analysis is a description of the patterns that occur within a data set and the overarching design that unites them. Boyatzis (1998) suggests that this description may take several forms. At one end of the spectrum it may result in a list of themes, at the other end a complex model with themes, indicators and qualifications that are causally linked, or alternatively anywhere between these two poles. A theme is described as “a pattern found in the information that at the minimum describes and organises possible observations, or at the maximum interprets aspects of a phenomena” (Boyatzis, 1998:vii). This may be identified at the manifest level i.e. directly observable in the data, or at the latent level i.e. underlying the phenomenon, and may initially be generated inductively from the data itself or deductively from theory and prior research. Although Green and Thorogood (2009) describe thematic analysis as an essentially comparative process, suggesting that it is the most basic type of qualitative analysis, Perakyla (2008) suggests that in research designs where the qualitative text analysis is not the core of the research, but instead is in a subsidiary or complimentary role, as in this study, no more sophisticated text analysis method may be required.

Thematic Analysis was used to analyse the interview transcripts in this study. The essential features of thematic analysis made it suitable and appropriate in that it does not rely on the specialised procedures of other means of qualitative analysis, and can be applied across a range of theoretical and epistemological approaches (Braun & Clarke, 2006; Schwandt, 2007). The thematic analysis method is particularly idiosyncratic and several processes of thematic analysis have been described (Boyatzis, 1998; Braun & Clarke, 2006; Schwandt, 2007; Grbich, 2007; Green & Thorogood, 2009). The study used the form of thematic analysis described by Braun and Clarke (2006) which is a recursive process involving six main stages [Figure 6.1].

Figure 6.1 *Stages of Thematic Analysis*

Stage	Phase	Description.
Stage 1	Familiarisation with the data.	Transcribe the data, read & re-read the data; note down initial themes.
Stage 2	Generating initial codes.	Code interesting features of the data in a systematic fashion across the entire data set; collate data relevant to each code.
Stage 3	Searching for themes.	Collate codes into potential themes; gather all data relevant to each potential theme.
Stage 4	Reviewing themes.	Check if themes work in relation to coded extracts [level 1] and entire data set [level 2]. Generate thematic map of analysis.
Stage 5	Defining and naming themes.	Ongoing analysis to refine the specifics of each theme and the overall analysis the story tells; generate clear definitions and names for each theme.
Stage 6	Producing the report.	Selection of compelling extract examples; final analysis of selected extracts. Relating back of the analysis to the research questions & literature. Producing report of the analysis.

(Braun & Clarke, 2006:87).

Braun and Clarke (2006) propose that the first phase of thematic analysis involves immersion of the researcher in the data to the extent that they are familiar with the breadth and depth of the content. This they suggest involves “repeated reading of the data in an active way” (p.87). As Gillham (2005) suggests that the transcription of interviews results in the loss of the semantic properties of human voice which can radically alter what the words mean, data immersion in this study involved the reading and rereading of interview transcripts while simultaneously listening to the audio recordings of the individual interviews. This procedure was followed in order that the nuances and paralanguage of the interviews was not lost, and the written accounts could be checked and rechecked for accuracy against the audio recordings. Initially the entire data set (i.e. all twelve interviews) was repeatedly studied as a whole with general ideas and impressions noted. This was followed by the repeated study of each individual interview transcript where again impressions and ideas were noted and then compared with those from the entire data set.

The second stage of analysis involved the generation of initial codes from the data. In qualitative inquiry a code refers to “a word or short phrase that symbolically assigns a summative, salient, essence-capturing, and / or evocative attribute for a portion of language based or visual data” (Saldana, 2009:3). Boyatzis (1998) suggests that a good code not only captures the qualitative richness of the phenomena but will also have the maximum

probability of producing high inter-rater reliability and validity. Interview transcripts were examined and relevant extracts from the transcripts were collated to form codes. Although this study was informed by Family Stress, Adaptation and Resiliency Theory (McCubbin & McCubbin, 1993; McCubbin *et al*, 1996), and interviewees were selected on the basis of IFS scores, the process of coding interviews was predominantly data driven. Codes were derived inductively from the raw interview transcripts. This procedure of empirically identifying codes was adopted in an effort to keep the data as close to the participants' experiences as possible (Gibson & Brown, 2009) and to increase the likelihood that others examining the raw data would perceive and encode the information similarly (Boyatzis, 1998).

The coding procedure was performed manually. Codes were identified, numbered, and then matched with data extracts that demonstrated the code. Codes were then categorised according to their focal area i.e. related to the child, the family or to the services. An example of this is presented in Figure 6.2. Initially 69 codes were identified; however additional refinement reduced this to 36 separate codes for which the inclusion criteria were clearly specified⁷. Ten of these codes related to issues associated with the child, fifteen to issues associated with the family, and the remaining eleven to the family's engagement with service. The codes pertaining to each of the three independent focal areas were then explored separately to identify the subthemes that related to each area.

Figure 6.2 *Example of Coding Strategy*

Code	Inclusion Criteria
03 – Uncertainty & Unpredictability.	This code relates to uncertainty relative to the child's condition. Included are references to the instability / unpredictability of the child's condition and the need for repeated hospital admissions. Descriptions of sudden medical crises are included.
Data Extract	
<p>"and I noticed that he was going blue and he was shaking a lot of the times and eh..... one of the times he went really blue and I phoned the GP and she said to me to immediately bring him in" [Interview 03]</p> <p>"we were in [hospital] and he was getting better.... the first initial visit and he was getting better and I thought "yea this is o.k. we can do this, we can manage this"and then he did this u-turn within a space of an hour..... and the next thing they're bringing him in and they're putting this cannula in his scalp which is....which was my lowest moment ever.....and all the panic.... [Interview 08]</p>	

Once all the data were coded and collated the third phase of data analysis involved the search for broad themes. Rossman and Rallis (2003:282) distinguish between a code which they

⁷ This coding framework was used for the assessment of interrater reliability previously described.

proposes is a word or phrase describing some segment of data that is explicit, and a theme which describes something more subtle and tacit. Braun and Clarke (2006:82) propose that a theme “captures something important about the data and represents some level of patterned response or meaning within the data set”. The subthemes from each area were reviewed in the context of the entire data corpus, and overarching themes identified. These main themes were then checked against each other, each subtheme, and against the notes and impressions originally documented during familiarization with the data corpus to ensure that they were consistent, coherent, and distinctive.

6.5.1 Ensuring the Rigor of the Analysis

While there are definite guidelines to navigate important threats to validity and reliability in qualitative research, there is no specific litmus test that can be applied that will confer a stamp of approval to a qualitative project (Wolcott, 1992; Nagy Hesse-Beiber & Leavy, 2010). Mays and Pope (1995, 2002) propose that the reliability of analysis of qualitative data can be enhanced by organizing an independent assessment of transcripts and comparing agreement between the raters. Boyatzis (1998) also proposes this process of establishing consistency of judgment as an important reliability check in qualitative research. Barbour (2001) concurs, suggesting the process of multiple coding concerns the same issue as the quantitative equivalent “inter-rater reliability”, and is a response to the charge of subjectivity sometimes leveled at the process of qualitative data analysis. Julien (2008) proposes an acceptable reliability coefficient in qualitative content analysis would reach a minimum of .60 (i.e. 60% agreement between different coders).

In the interest of validity and reliability 25% of the twelve interview transcripts were subjected to independent verification by a second coder (n=3). The second coder, who was entirely independent of the research project, received the coding framework and three randomly selected interview transcripts, and was asked to code the transcripts using the coding framework provided. The coding framework indicated the codes and their defining attributes. No additional information was supplied. The results were then compared with those of the primary coder. Agreement on individual codes ranged from 75-100%, with an overall agreement of 95.6%. When the framework was applied to the individual interview

transcript coding agreement ranged from 89.6% to 97.6%, with an overall inter-rater reliability of 94.4% on the transcripts [Appendix S].

Although Lincoln and Guba (1985) propose that member checking is essential to the credibility of the qualitative research project it was not employed as a quality measure in this study. More recently there is debate about the value of this process as a validation strategy in qualitative research with several commentators questioning its universal appropriateness. Mays and Pope (1995, 2000) identify several potential problems. They suggest that if used in isolation this technique is based upon the assumption that fidelity to the participants' common-sense perceptions is the touchstone for reality. They also suggest that apparently discrepant views are likely since the researcher produces a broad and holistic account designed for a wide audience which may be different from the account of an individual informant simply because of their different roles in the research process. Additionally Barbour (1998) cautions that participant validation exercises make considerable demands on participants' time and, depending upon the research topic and the content of transcripts, can potentially be distressing or even exploitative.

The decision not to seek respondent validation was made in the context of several issues pertinent to the study: interviewees were chosen on the basis that they had already participated in the survey used in Stage One which made it possible to triangulate the findings with those from the previous stage. All of the interviewees had already been subjected to a double burden of participation (survey and interview), and additionally, given the nature of the study, had extremely busy and complex home lives with many demands on their time. Given the high inter-rater reliability that had been achieved it was considered that seeking respondent validation would impose an unnecessary additional burden on participants for relatively little additional benefit, and could be exploitative of the good will that participants had already extended towards the study.

6.6 Description of Interview Participants

All of the interview participants in this phase of the study were mothers. The child's father was present during one interview but he did not participate in the actual interview process.

All geographical areas were represented, with interviews conducted in Munster, Leinster and Connaught, in both urban and rural areas.

Interviewees ranged in age from 23 to 44 years (\bar{x} =34, SD=6.73). Two interviewees were single parents (17%), and 92% worked full time in the home caring for their child (n=11). Three mothers had no other children, six had one other child, and the remaining three mothers had two, three and four children respectively.

The children with a life-limiting neurodevelopmental disability ranged in age from one to six years (\bar{x} =3.3, SD=1.7). One child's diagnosis was unknown, five had an organic central nervous system disorder while the remaining five had a chromosomal abnormality. All children were involved with a variety of services and health professionals (\bar{x} =8.5, SD=2.5) including both general paediatric and intellectual disability services. Two children had already been referred to specialist palliative care services. The mean number of health related problems experienced by the children was 6.5 (SD =1.98), ten of the twelve children were dependent on some form of medical technology.

The previous sections of this chapter have presented the process of selecting participants for interview, and the interview process. Data analysis was discussed, and the strategy employed to ensure rigor of the analysis presented. A description of participants' demographic data has been presented to place the findings of the interview analysis in context. Although each family who participated in this study had a unique experience, when these experiences were compared certain commonalities emerged. The following section presents the results of the thematic analysis of mothers' interviews.

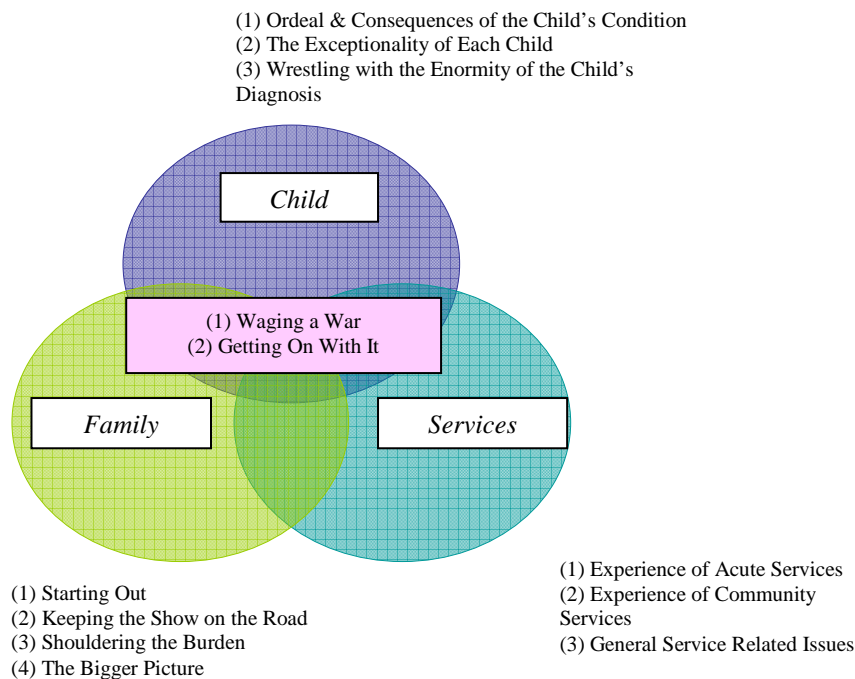
6.7 Findings

The results of the thematic analysis are presented in two stages. Initially the themes related to the focal areas of the study are presented (the child; the family; and the experience of engagement with services). Presenting these themes fulfils the function of explaining and augmenting the findings from the parents' surveys which was the primary purpose of this stage of the study. Themes are supported with text segments from the interview transcript. These text segments present interviewees verbatim responses, sometimes demonstrating the

frustration that mothers experienced. Italics are used in quotations to represent the emphasis mothers placed on particular words or phrases during the interview. Hesitancies and pauses in speech are included.

The second stage of the findings presents the super ordinate themes that overarched the entire data set. Presenting these super ordinate themes facilitates a more holistic and three-dimensional understanding of the multi-faceted nature of family life and engagement with services of young children with life-limiting neurodevelopmental disabilities and their families. The relationship between the themes and super ordinate themes is presented in figure 6.3. The chapter concludes with preliminary discussion of the findings and the main conclusions that can be drawn from this stage.

Figure 6.3 *Themes and Super Ordinate Themes from Mothers' Narratives*



6.7.1 Child Related Themes

Three themes emerged in relation to the young child with a life-limiting neurodevelopmental disability: the first related to mothers' descriptions of the ordeal of the child's condition; the second to the individuality and uniqueness of each child; and the third to the enormity of the child's diagnosis.

6.7.1.2 The Ordeal and Consequences of the Child's Condition

This theme relates to the difficulties and challenges that the child experienced and their consequences for the child. It incorporates mothers' descriptions of the physical and social challenges that the child endures; the unpredictable nature of the child's health status; and the unrelenting and repetitive nature of the care required.

The children in this stage of the study had rare conditions which resulted in multiple, complex and on-going health problems. Most of these problems were present from birth or shortly thereafter, and many required complicated medical and technological management. This resulted in mothers carrying out highly skilled nursing care in the form of complex drug regimes and the use of specialised medical equipment (most frequently to support gastrointestinal and respiratory function). However the children also required complex care in another sense. In addition to the procedural knowledge which allowed mothers to provide high levels of specialised care, mothers expressed a form of tacit knowledge which allowed them to almost instinctively know if the child was developing a problem and ensured they were totally attuned to all aspects of their child's needs. One mother commented

“she is visually impaired , she is deaf, she has Cerebral Palsy, she has oesophageal varices.....she has major liver problems and her day to day as in from getting up in the morning to getting in the routine.....she will feel the vibrations on the floor and it lets her know that you are coming in, she will feel vibrations and she will smile.... but if you just went in and opened the cot and leaned over she will get the *fright of her life*.... like if you just touch her, so you have to go in slowly and move around and let her know that you are there.... then she will just start coming around, she is very nervous..... and it is not nervous that she is afraid, she is nervous that she does not know what is coming next really.... because her senses would be *very bad*....yeah.... she is peg fed, she wears hearing aids....and eh....no, she can get break through, so she gets her MST which is the big one... if she is having a *bad* day she would say..... well you would know by looking at her , you would know just by her *face*.. you would *know* she is having an off day” [35: mother of child aged 4]

The children's health status was unstable and unpredictable, often involving a "revolving-door" situation with acute hospital services. Frequent hospital admissions were seen as a source of suffering for the child as each time they were subjected to the necessary routine of investigations and interventions. In many cases the nature of the care required at home on an ongoing basis was unrelenting and repetitive. Some children required around-the-clock care whereby drug regimes or feeding schedules needed to be continued and attended to during the night. Maintaining this level of care necessitated developing and adhering to a strict routine. Feeding and drug schedules all needed to be performed at pre-determined times. Between these times the other care requirements of the child needed attention. Meticulous planning and timing of care was essential. This daily cycle of care was obvious in mothers' descriptions of how preparing and arranging the following days care marked the conclusion of the care required each day. Adhering to this strict routine was essential as the consequences of failing to be prepared, or making an omission, could cause the child to suffer or have severe consequences for the child's health status. The instability of the child's condition had consequences beyond the need for an exacting routine. One mother described this constant state of flux in the child's condition as the most difficult thing to cope with

"the *uncertainty* as to when he is getting sick next....and that's it....just having to be *always* prepared for that, and not knowing when it is...." [16: mother of child aged 2].

Many others commented on the practical difficulties that arise as a consequence of the child's unstable condition. These included never being able to leave the child unattended, not being able to leave the house unaccompanied, and the difficulties of trying to travel with the child

"and I remember being in an *awful* fret one of the times....I was in the car and I was on the motorway and [child] decided to have a seizure....and I had to stop the car and I had to go and apply the Oxygen... I had to apply the Stesolid....and there was a *huge* traffic of people behind me, and I remember they were all just beeping....and I remember it *very* vividly, even to this extent...I remember afterwards, after the seizure....getting back into the driving seat....and all these people passed me, and they looked through the window staring at me as like "did you not know that you are holding up the traffic"..... I remember just sitting there you know.... for half an hour....and I cried my eyes out....[04: mother of child aged 4 ½]

These unpredictable and ongoing health problems also resulted in negative social consequences for the child. The cause for this appeared to be twofold. Some mothers

described what they felt was the exclusion, by others, of their child from normal social situations and everyday events, while others made the decision to insulate the child themselves based upon their interpretation of others reactions

6.7.1.3 The Exceptionality of Each Child

Despite the complex difficulties experienced by the child, and the associated level of care required, this was not the main frame of reference for mothers when speaking about their children. This theme relates to mothers' descriptions of the individuality, uniqueness and "specialness" of their children, including the positive benefits to other family members that the child has provided. A strong emphasis was placed on this by mothers during the interviews.

Despite the difficulties and negative consequences of the child's condition every mother spoke emphatically and lovingly about the individuality and exceptionality of their own child. This took the form of expressions of love and devotion, and emphasis on the unique personality traits of the child, and a focus on the child's positive achievements. Mothers were anxious to point out the unique personality traits and quirks of the child: the things the child seemed to enjoy, and those they did not, the ways in which they interacted with others in the family, the ways in which they could make themselves heard even if unable to vocalize. Mothers described all the things which differentiated their young child with a life-limiting neurodevelopmental disability from other children and marked them as unique and individual. Many mothers attributed qualities of strength and courage to their child, particularly in the context of the prolonged suffering associated with protracted hospitalisation, medical tests and interventions. The child was frequently referred to as "a fighter" who had defied medical prediction of their longevity, achievements and capabilities. This concept of defying the odds appeared to be interpreted as a positive sign relative to the child's prognosis

"she's defying the odds *all* the time.....that you know, you kinda say "well they were *wrong*"...you know last year they told me she had three months to live... [35: mother of child aged 4]

Mothers were also anxious to emphasise the positive advances and achievements that the child had made. These were a significant source of celebration and pride, no matter how small.

It was apparent from mothers' narratives that, from the outset, the child was perceived as central to the family unit. Within the family the child was seen to make a significant positive contribution to both their mothers' personal development and, where present, to the development of siblings. Mothers spoke of their child in a transformative way, contributing to their own positive personal growth. They believed that, as a result of their child, they were now less concerned with superficial aspects of life, had more tolerance, were stronger and more assertive, and had increased empathy towards others

"I am *such* a different person now... I have more patience, I have more understanding, I have more *compassion*..... I never knew what... a special family before..... and never stopped to *think* what their lives would be.... *now* I stop and I think.....[04: mother of child aged 4 ½]

Mothers also proposed that having the child as a member of the family accrued benefits for other siblings where present. However, unlike the mothers' benefits, these were generally intangible. So while mothers made reference to the fact that the situation could only benefit other children in the family, they were less explicit about exactly what these benefits to siblings were.

6.7.1.4 Wrestling with the Enormity of the Child's Condition

The final theme related to the child was associated with mothers' description of their attempts to grapple with the child's condition and its potential implications for the future. Some children's difficulties were obvious from birth, for others diagnosis took some time. Mothers reported conflicting perceptions of this delay: for some mothers a delayed diagnosis caused considerable anxiety both in terms of the uncertainty of what to expect, and the potential consequences of the delay for the child's treatment; for other mothers a delay seemed to provide a form of respite, a method of avoiding a potentially unpleasant reality

"on one hand it would make things difficult because you don't know what to expect.... but then on the *other* hand like.....maybe it's *not* such a bad thing, you know.....not knowing.... [12: mother of child aged 1].

In situations where specific diagnosis-related information was provided mothers described the painful challenge of trying to assimilate this. Others were left to try to obtain information of the child's condition themselves

"I just felt my *world* had collapsed around me, when I heard "brain" I just said "oh my God" didn't know *what* it meant, when the nurses wrote down the actual name and said to me "well there it is if you want to get some of your family members to look it up for you" did that and well... that was a *bad* idea..... that was a *really, really* bad idea....." [31: mother of girl aged 5]

Accepting the child's diagnosis and its implications presented a long struggle for mothers, and this was not always a straightforward process. For some there appeared to be a gradual acceptance of the child's reality, while others appeared to alternate between knowledge and denial

"she said "there's a 50% chance that he won't make it through" and I'm kind of saying "*no*....that's *other* babies..... that are worse off than [child]"because you know you don't let yourself believe it..... how actually bad he *is*..... because if you did you just wouldn't get through[16: mother of child aged 3]

Only one mother made explicit reference to the life-limiting element of her child's condition during the course of the interviews (although this did not prevent her making plans for the child's future). This particular little girl was in the terminal stage of her illness and had already been referred to specialist palliative care services. One other child had also been referred to specialist palliative care but this was perceived as a negative development by the mother who strongly refuted the need for the service at this time. There was a sense that accepting palliative care for the child was an admission of defeat particularly because it conflicted with the parents' own goals for their child

".... and I struggle... all of the time ever since I was involved with them, or put in touch with them, I *battle*.... I have had meetings with the... I have had *arguments* with them, and it's a mind set that I can't change.... *they* want to treat her for quality of life and comfort and I *don't* want to.... I know that they think myself and my husband are ridiculous....to even *think* that [child] might ever even sit....and that's something that is a real goal for us.....and even if never happens.... we have to have this goal in our heads, because otherwise we're doing *nothing* for her.... and I feel... as a parent.... well that I wouldn't be much of a parent if that's what I was doing.....and she is such a *great* little girl....I think it's a shame not to want more for her..." [34: mother of child aged 2]

Not only was the life-limiting element of the child's condition not alluded to by most mothers, but all of the mothers in this phase of the study spoke of the plans they had for the child's future. This was true even of the mother whose child was in receipt of palliative care services. Most mothers spoke of always retaining a glimmer of hope, the sense that the outlook for the child may change because "you can never be sure what's around the corner". This allowed some mothers to make long terms plans for the child

"in my mind like [child] is going to grow up to be a man..... and we are going to plant a few Christmas trees down for him..... and we are going to build him a mushroom tunnel and do vegetables.... and I have *great* plans for him you know.... that he can be chopping the firewood and things like that....." [16: mother of child aged 3]

6.7.1.5 Interim Summary of Child Related Themes

These themes portray a picture of the physical and social challenges experienced by the child with a life-limiting neurodevelopmental disability, and an insight into the complex and often unremitting nature of the care required. However these difficulties were not the main frame of reference for mothers who did not seem to view this burden of care separate from the love and devotion they felt for the child, and the positive benefits they believed the child brought. Coming to grips with the child's condition was a protracted and difficult process, and in many instances a conflicted one. Despite the numerous and severe complications the children experienced, and the level of on-going care they required, the majority of mothers did not make reference to the life-limiting aspect of the child's condition, nor did this appear to inhibit making plans for the child's future.

6.7.2 Family Related Themes

The impact on the family of caring for a child with a life-limiting neurodevelopmental disability has been quantified in Chapter Six. This section presents the themes that relate to mothers' descriptions of the management of everyday family life and relationships when a child has a life-limiting neurodevelopmental disability, and how the family manages to continue to function and survive.

Four themes were identified relative to the concept of family life. The first of these "Starting Out" relates to mothers' experiences of the birth of their child and the issues associated with

coming home with a life-limited neurodevelopmental disabled infant. “Keeping the Show on the Road” described the strategies families employ to manage life day-to-day and the resources that they draw on to do this. The third theme “Shouldering the Burden” describes the impact that having a child with a life-limiting neurodevelopmental disability has on the individual members of the family, and its impact on the relationship between the mother and father. The final theme “The Bigger Picture” relates to the world outside the family and how this too must be navigated and managed.

6.7.2.1 Starting Out

This subtheme relates to mothers experiences of the birth of their child and associated issues. All mothers began at this point, the beginning of their journey, and told their story as a temporal narrative from this point. The most minute details were remembered with exactitude. Dates and even times were recalled with ease, as was each individual they encountered in both positive and negative contexts.

For some mothers the fact that there was a difficulty with their child was obvious immediately, but those for whom this was not the case expressed that a major difficulty at the outset was being taken seriously when they expressed their concern that something was “not quite right” to hospital staff. Concerns were only accepted as legitimate when the problem was witnessed and corroborated by a member of the medical or nursing team. This left mothers feeling vulnerable and full of self-doubt with regards to their ability to recognise symptoms.

“and it was just like I was not believed..... it was like my word wasn’t anything... what *I* saw, you know...*they* didn’t witness a seizure so they sent me back and they said to me that it was probably regular childhood movements..... even though I think.... that at this stage you know, I have raised three other kids...[04: mother of child aged 4 ½]

When it was established there was a problem with the child other difficulties ensued which left mothers feeling alienated, powerless and lacking a sense of control. Some of the mothers reported that their baby needed transfer to another hospital creating a physical distance between them and their infant; others described the difficulties of having a child in an intensive care unit even when this was in the same hospital as the mother. The lack of

opportunity to physically interact with the child was a cause of distress, and some mothers expressed that it deprived them of the opportunity to “get to know” their infant. While in some circumstances lack of physical contact was the inevitable result of interventions, monitors and investigation, in others the cause was less obvious to mothers who associated it with the rigid rules and routines of the special care unit, in particular the lack of visitation rights. One mother described how her infant developed difficulties and was taken to special care just hours before they were due to be discharged from hospital, and how not only was she expected not to accompany the child, but was instead expected to get ready to go home.

Many mothers’ reported difficulties associated with the communication of information about their child’s condition. In some situations, especially where the child’s condition was rare, mothers felt that there was not enough information available to them, either because medical staff did not have the information, or because they were reluctant to speculate about the prognosis for conditions with which they had little experience. Mothers described being given distressing information when they were alone and unsupported, or described how they simply could not make sense of the information provided

“the pediatrician explained about the two sections of the brain, and about this group of cells in the centre and they just hadn’t developed.... he never *ever* mentioned the word disability at all.... *Never*.... didn’t...I was very upset when I heard brain or whatever....I was very upset, and to be honest with you, if he asked me what exactly did he say I probably can’t remember all of itbut I just remember him when he said about the brain..... as a matter of fact I actually remember thinking *oh my God* that means he said the group of cells that connect the brains hasn’t developed..... I just had pictures of her brain wobbling in her head....To me it was like oh my God what happens if she moves her head.....it was all just completely and utterly *shock*....[husband] wasn’t with me when I was told.....[31: mother of child aged 5]

Many of the children required a protracted initial hospital stay. While some mothers chose to remain with their child, others made a daily commute to the hospital, often from considerable distances away. Both the general environment of the hospital and lack of basic facilities caused considerable difficulties for mothers. In particular sleeping facilities for parents were a significant problem for parents who stayed with their ill child

“the little stupid things like.... the likes of the hospital and stuff you have the little skinny mattresses....I mean would *you* sleep on that for seven nights a week... you are making

yourself *sick* with these services that people don't realise..... the little small things, like a mattress that size you say to yourself "you try and sleep on that"....." [35: mother of child aged 4].

Once the child's condition stabilized mothers faced being discharged with their infant. Without exception all mothers felt totally unprepared for this. Most expressed that they felt scared and out of their depth. There was the perception that this enormous crisis of confidence was either ignored, or trivialized, by hospital staff. Mothers used adjectives such as feeling "scared", "overwhelmed", "traumatized" and "demented" to describe their feelings on discharge from hospital. They felt they lacked both the skills and knowledge required to care for their infant. They expressed that they did not have sufficient procedural skill to care for the child particularly in relation to children requiring technological assistance, and also felt that they were not adequately informed of what to look out for in terms of changes or deterioration in the child's condition. It seemed to mothers that they were left to their own devices with no information except the information they could gather themselves

"we hadn't a *clue*....and she said "right, off you go", and we were like "how do we *feed* him", and she said "just do as you normally do"..... *Jesus*, sure we hadn't a clue..." [13: mother of child aged 1 ½]

In addition, mothers and children were often discharged without pre-arranged local support or knowing who to call on if difficulties arose. For some mothers, particularly those from rural areas whose child had been transferred to Dublin, telephone support from the referral hospital was all that was available, and this was inconsistent. Many mothers described being totally overwhelmed by the constant care requirements of the child and lack of support

"There was times I just cried *all day everyday*..... there was times when I could not honestly get up off the chair to go to the *toilet* because she was so bad.... [child] was so bad that when you would leave her down she would just go hysterical..... that you couldn't even go to the toilet....you know....but there was times.... like I often had a day I just had toast or a bit of bread and that would be it for the whole day..... because you wouldn't have *time* to make anything.... so you're eating and you're picking and you're grabbing.....when you got 5 minutes you know...all the time watching the door for someone to come and help...." [61: mother of child aged 2 ½]

For most mothers this state of crisis eventually receded, and a state of semi- equilibrium evolved. This was not always quick to develop and appeared to be contingent upon the support (social and services) that mothers could draw down. Mothers became less fearful of the technical aspects of care and more confident in their ability to provide the care their child needed. At this stage the focus appeared to shift towards attempts to construct a framework or routine in which day-to-day care could be continued in the longer term.

6.7.2.2 Keeping the Show on the Road

This theme relates to the family's attempts to manage day-to-day life and reconstruct a sense of order from the fragmentation produced by the child's condition. In order to achieve this family life needed to be planned and executed with military precision. The development of a structured family routine was essential. This was particularly important in families where there were other siblings. The routine was structured around fitting the family in with the child's needs and schedule, and seemed only to vary in accordance to the particular appointments the child had on any given day, or unanticipated alterations in the child's condition. All family plans were provisional and subject to change depending upon the kind of day the child was having.

Strategic planning and routine was also required to overcome the practical difficulties that were encountered on a daily basis. Driving was a major issue for mothers, both in terms of the amount of equipment that needed to accompany the child, and the fact that in many cases the mother could not travel alone in the car with the child. This meant that trips needed to be scheduled in advance so that arrangements could be made to have accompaniment. In some cases the child's requirement for constant care also meant that even normal household chores had to be scheduled in advance in order that they could be performed when assistance was available

“and Monday to Thursday when I have [carer] for the 4 hours in the morning I'm trying to get everything done.....'cause God knows you're not going to get a chance to do it on Friday, Saturday or Sunday....[61: mother of child aged 2 ½]

One of the major issues that needed to be addressed in order to manage the practicalities of day-to-day life was the demarcation of roles between parents. Without exception all of the mothers were the main providers of care and while in some situations aspects of the physical

tasks of care were shared, the main responsibility of overall care management fell to the mother. In some families this appeared to be a negotiated and pragmatic arrangement, with mothers describing that they assumed this role because they no longer worked outside the home. In other families mothers appeared to have assumed the role by default

“we don’t disagree, No..... not really.... because [husband] would tend to leave it a lot to me.....and [husband] made an announcement that he doesn’t actually *like* hospitals anymore.... and I’m kind of going “yea, like I find it a barrel of laughs”..... [16: mother of child aged 3]

When both partners worked in outside employment it was the mother who took time off if the child was ill or hospitalized, and who organised hospital appointments and tried to arrange services for the child.

Mothers drew on several sources of practical and psychological support to assist them in the day-to-day management of family life. In general family and friends could be called on to provide psychological support although mothers were more hesitant to request practical assistance with care. In many cases the level of specialised care required by the child precluded obtaining help with childcare from family and friends. Some mothers felt that there was a limit to the extent that they could call on practical assistance from family and friends. In these situations services became an increasingly important coping resource, with one mother whose child required constant care describing it as a “relief” when the child was hospitalized

“it’s a *relief* when [child] goes into the hospital I have to say..... I know it’s an *awful* thing to say.....but it’s relief to be quite honest..... and I mean I stay all day with her, I put her to sleep and I go home... but the fact that you are going home and getting into bed and at least your mind is shutting down.....[61: mother of child aged 2 ½]

Mothers also turned to other parents in situations similar to their own as a source of support. Other parents were considered a particularly important source of information and advice on services and entitlements. Some mothers developed new skills to help them manage, actively working to become more assertive and knowledgeable, and often undertaking courses that they thought would benefit the child now or in the future.

Throughout all of the challenges encountered day to day families struggled to maintain a sense of normality for other siblings, and to normalize their family situation. Mothers

described how important it was that the young child with the life-limiting neurodevelopmental disability was treated as normally as possible, both by the family, and those outside the family unit. At the same time mothers struggled with the task of reconciling the particular needs of this child with the general needs of other children in the family. Some mothers described this as one of the most difficult challenges they faced day to day, and many reported that the real value of having home nursing hours was the opportunity it afforded to spend time with other children

“the hardest thing is how to get through each day and give them *each* something...I mean its really kind of the *emotional* thing.... *that* kind of thing...it’s not the medical thing it’s....*that*” [25: mother of child aged 6]

This struggle to develop a sense of normality also appeared to involve a framing of the family situation as a normal, or at least a not very unique one. Mothers described how in many ways their family was really no different from every ordinary, average, family. This perception appeared to be reinforced by availing of opportunities to meet other families in similar situations. Mothers actively sought out such encounters which reinforced the perception that neither the family situation, nor the challenges encountered, was unique. For many this involved attending family days organised by a service or charity with which they were involved. Others attempted to locate families similar to their own through their main services

“you actually look forward to them days...because you meet people that are the same as you....everybody there is the *same*....so it’s like you are in with the crowd....[35: mother of child aged 4]

Mothers described two particular approaches that helped them to keep going day to day. The first appeared to be a combination of stoicism and pragmatism, and the second a deliberate focus on accentuating the positive elements of the situation, always remembering that things could be worse. Mothers suggested that they managed simply because there was no alternative; it was just what they had to do

“sure I *have* to keep going....because if I don’t what’s going to happen then you know....you just have to keep going....” [12: mother of child aged 1]

The difficulties and challenges of the situation appeared to be made easier by deliberately focusing on positive aspects and remembering that, no matter what the circumstances, there were other children and families in worse situations

“[condition] can be quite profound in some children..... so [child] really could be a *lot* sicker than she is....I mean she could be PEG fed, she could have problems with aspiration and she doesn’t...well I mean....you know she does.... aspirate *fluids*...but I suppose what I’m trying to say is that she could be a *hell* of a lot worse than she is... so I feel blessed that I am not dealing with that level of care..... [34: mother of child aged 2]

Mothers also strived to make sense, or meaning, of their situation. For some spirituality was important as they sought what they described as a “sign”, while other mothers achieved positive meaning through “giving some thing back”. This concept of reciprocity was operationalised in a variety of ways. Some mothers felt it was important to act as an informal resource to other parents, others collected for charities or participated in events to raise funds. Some became politicized and acted as political advocates for children and families in situations similar to their own by volunteering on boards of management and advocacy groups.

6.7.2.3 Shouldering the Burden

This theme relates to the specific impact having a child with a life-limiting disability has on individual members of the family, family relationships and the family unit. For although mothers were anxious to elaborate on the positive elements of having a young child with a life-limiting neurodevelopmental disability they also described a range of negative physical, psychological and social consequences of the situation for themselves and for other members of the family.

Some mothers described that the care requirements of the child were sufficiently burdensome and time consuming that there was little time left for basic self-care. This left some feeling despondent and overwhelmed

“I am *literally* under house arrest like if we you’d be constantly ringing people like “can you go to the shop for me?” or “can you drop me in something to eat?”.... like and some days me Ma would drop in and say “did you have anything to eat today?”... and like I could literally sit down and *cry*..... like how can I have anything to fucking eat, *look* at me look, at

the *state* of me...my house... look atsome days she would not sleep at night so you are up all night and you are up the next day...” [35: mother of child aged 4]

Many also found the care required by their child took a physical toll. Most frequently mothers described musculoskeletal problems associated with the effort of lifting and carrying (either the child as they got bigger and heavier, or the equipment necessary for the child’s care). Chronic exhaustion also appeared to be a pervasive problem. A persistent state of tiredness and fatigue was reported by all mothers which was generally described as feeling “physically and emotionally drained”. For many this was attributed to a chronic lack of sleep associated with some medical intervention the child required overnight. However, mothers reported that they found it almost impossible to have a restful night even when no overnight intervention was required

“at night time now I am half awake and half asleep..... because I don’t *know* if she is going to start writhing and choking on the vomit... ya I am..... always *one eye open*.... yes... [61: mother of child aged 2 ½]

Despite this night time respite was extremely limited, and in cases where in-home night respite was available mothers reported that the more complex the child’s needs the more difficult it became to find a suitable carer. Some mothers reported that they felt guilty for asking their partner to share night-time care, particularly if the partner had to get up in the morning.

Although some mothers had in-home support during the day this did not alleviate the problem of tiredness. In-home support simply allowed mothers to attend to the needs of other children, or perform the ordinary everyday household chores that are part of family life and which could not be performed if scheduled help was not available.

“I suppose the hours that I would get from [service] I would tend to use for practical purposes.... like going and doing grocery shopping, or getting the washing done you know..... I have *never* used them for myself.....” [39: mother of child aged 4 ½]

Despite a backdrop of constant fatigue and exhaustion mothers described the need to remain constantly alert for changes in the child’s condition. These could happen suddenly and without warning. The child’s rollercoaster health status appeared to be a cause of constant tension for mothers who needed to be constantly alert and responsive to the ever changing

fluctuations in the child's health. It also meant that mothers had to be permanently available in case a difficulty arose with the child when the mother was not physically present. They were aware that they could be called upon at any time to attend to a difficulty with the child.

Social isolation was a frequently encountered problem for mothers. Some had left paid employment to care for their child and many missed the social opportunities that work outside the home had presented. Others had little opportunity for social intercourse due to the burden of care required by their child and mothers described the difficulty of maintaining friendships when friends don't seem to understand the complexities of their life.

Mothers also described the constant worry and anxiety they experience about the short and long term future and the potential challenges that might occur. This anxiety appeared to be focused on two main areas. The first related to services, with mothers describing the constant worry of future service withdrawal⁸. The second related to what the future held for their child in the long term. Although one mother described how the future was just too difficult to contemplate in its entirety at this time

“....and I don't want to see a child say older than 5 with this condition..... because I just need to focus on what's going on at the moment..... I got the DVD on the first 18 months.....and you are allowed it for a month or something..... and after 3 weeks I *still* hadn't watched it because I was too scared to watch it.....and I was afraid that I am going to put this on now and there will be something on it that I know or I haven't realised so farand I don't want to know about it and I am going to be back down to square one again.....[16: mother of child aged 3]

Potential service reduction appeared to be a constant cause of anxiety for the mothers in this study. Every mother was aware of, and frequently referred to “cutbacks”: a reduction in services and staff associated with reduced budgets and funding. Many mothers worried that particular services would not be available when their child needed it. Where children were currently in receipt of services mothers worried that this would be withdrawn, or reduced, in the future

⁸ This was a particular challenge in relation to the withdrawal of a national charity service which generally supports families of children up to the age of four. This had been extended for all children over this age in this phase of the study.

“my *biggest* worry I suppose is ensuring his needs in the future are met.... and I just don’t feel that that is going to be placed based on the way things have moved at the moment [39: mother of child aged 4 ½]

In the face of potential service reduction, mothers also worried about their ability to continue to provide care for their child into the future, particularly as the child got bigger and more difficult to manage. More general worries about the child’s future were also described. Principally these were related to who would care for the child if anything happened to the mother. For the most part mothers hoped that this would fall to siblings or family members.

Another cause of anxiety for mothers was the issue of having another baby. While this was difficult for all mothers it was particularly problematic for mothers where there was a known genetic component to the child’s condition. The issue of genetic testing was complex, and did little to alleviate these worries. One mother, who knew she was a carrier of the child’s condition, described the difficulties associated with being pregnant

“Its very sad actually.... I *know* that I am a carrier of [child’s] condition right.... I carry it.... so I know that should I have any more children there would be a high possibility that they would be born like [child].... now to be honest I would have been told initially 18% but it is actually 42% chance....but we decided to have [child] anyway....(long pause)....

Interviewer: Was that difficult [name]?

“I suppose it was because I didn’t know what way he would be born you know.... I could *guess* that he maybe hopefully he might look a little bit like my brother you know..... and there would be nothing very *disturbing* wrong with him you know.... *visually* disturbing wrong with him..... and thankfully there wasn’t... but I suppose at some point we were kind of going “oh Jesus, what if he has no... missing fingers or whatever”.... [39: mother of child aged 4 ½]

Having genetic testing after the child was born brought its own set of fears over the decisions that might accompany the results

“I wouldn’t like to have to make that decision then..... *knowing* that your baby was sick..... You know.....do you or don’t you..... But like I could *never* look [child] in the eye again if I.... now everybody’s decision is completely an utterly personal, and I don’t blame *anybody* for any decision that they are forced into..... but I couldn’t look [child] in the

eye again if I had got rid of a baby that was not perfect..... I *couldn't* do that.... but do I have the energy to do it *again* I really just don't know..... [16: mother of child aged 3]

An additional feature of the mothers' accounts was a profound sense of loss and yearning. Mothers described personal losses they incurred as a consequence of the situation: loss of confidence and self-esteem, loss of opportunities for social interaction and employment, and the loss associated with changed status. They also described more intangible losses, in particular the loss of a "normal" child and the things anticipated and associated with normal childhood

"I *would* like to have the normal child.... the homework, the giving out, school,...you know, "put your shoes, on do this do that".... because you don't have that with [child] you are doing it all for her,... and to have a child and to have a little girl you always wanted you always wanted to *do* things, but to be told that they are not going to be... and to be hit like that.... but you know she just might be.... you always have that glimmer of hope.... [35: mother of child aged 4]

Mothers were not the only family members to incur a negative impact from the situation. Despite the fact that mothers struggled to minimise the impact on siblings, and actively worked to keep life as normal as possible for them, they also described negative consequences for siblings of the family situation. Mothers described how the burden of care required by the young child with a life-limiting neurodevelopmental disability often negatively impacted on the time available to other children in the family. This resulted in siblings not receiving the attention that mothers believed they needed which was frequently a source of guilt for mothers. At its most extreme, one mother described the situation in the following way

"[sibling] has *no* life you know.... she's way behind in her development I know that.... and I know being with [child] all the time doesn't help.... but I *can't* sit and do these learning jigsaws.....I just don't have the *time*..." [61: mother of child aged 2 ½]

Other negative impacts were also experienced by siblings. The unpredictable nature of the child's condition made planning family occasions almost impossible, and all family plans were subject to change at short notice depending upon the child's health status on any given day. Even scheduled activities and hobbies for other children in the family could be difficult

to organise and maintain in the face of the child's precarious health status. In addition frequent episodes of hospitalisation required by the child resulted in mothers spending frequent, and sometimes relatively long, periods away from home which some mothers felt was difficult for siblings, especially young siblings, to understand.

Like mothers, siblings too could be socially isolated as a consequence of the family situation. In some instances the fact that the child did not travel well limited family opportunities to enjoy time together away from the home environment. Often the complex and unpredictable nature of the child's condition eliminated the possibility of a family holiday as mothers were reluctant to take sick children too far from a hospital in which they were known. Sometimes siblings were not invited to friend's homes because of the limited possibility that this would be reciprocated

“you know I have been in situations where parents have come up to me and wonder why I don't do play dates..... I have had a teacher who asked would my child..... when my child was having problems in school and I went to speak to her and I said “why is my child having these problems?” and she said to me “I notice that you don't do play dates”and I said “I don't know if you know about my situation but I have a child who is in and out of hospital a lot..... I don't want to take the responsibility of somebody else's child when I have my child there” I said “I *can't* take the responsibility of somebody else's child”.... and she said to me “if you did these play dates the other parents reciprocate” I am wondering to myself *why does it take that* they all know of my situation you know..... but yet because I can't my children have to suffer.... [04: mother of child aged 4 ½].

More general impacts on the family were also described. In some cases the family home was overtaken by the equipment needed to provide care for the child leaving little space to accommodate other family members. Mothers also described financial strains associated with the care of a young child with a life-limiting neurodevelopmental disability. Extra money was required for episodes of hospitalisation (travel, food accommodation), for medications that were not covered by a GMS card, for equipment that was not supplied by services (or was too slow arriving and needed to be purchased), for home modifications that were not covered by grants, and in some instances to pay for private therapy (particularly physiotherapy) where the service fell far short of the child's needs.

In some cases the relationship between parents was also impacted by the child's condition and its associations

"I *definitely* think having a child with disability and a lot of medical needs does effect your relationship....*hugely*..... and even though [husband] is the best in the world like he really is..... it does *definitely* effect..... and I definitely think it can be one of... I think that probably it is a hard part as well..... [31: mother of child aged 5]

While many mothers described their partner as their greatest support in relation to the care of their child, the situation did not appear to be without consequences for other aspects of the couples' relationship. Socially parents had little opportunity to spend time together. Many mothers described that they rarely, or never, spent social time with their partner. For many this time was just not available, for others the complexity of the child's condition made getting out impossible as no suitable childcare could be arranged. Intimacy too could suffer as a consequence of the situation. Some mothers felt that they were unable to leave the child unattended overnight. Often this resulted in parents sharing a room with the young child or the mother sleeping in the young child's room.

It appeared that relationships could suffer in other ways too. One mother described the difficulty of trying to communicate, and the isolation experienced, when both parents do not have the same goals for the child. There was one mother who lived alone who participated in the interview stage of the study. This mother described how the constant care requirements of the child made it impossible to sustain a relationship, and how cautious she would be about introducing a new potential partner to the situation.

6.7.2.4 The Bigger Picture

In addition to the challenges experienced within the family mothers also described challenges encountered in the world outside. While mothers could recount specific incidences which were positive in terms of individuals providing assistance in difficult circumstances, most described an outside world characterized by a general lack of sensitivity and understanding.

Mothers reported that often people were slow to provide help or assistance in social situations, even when they were obviously struggling with the child or the child's equipment. They also frequently described what they believed to be social perceptions of their child as a

curiosity. This was encountered almost on a daily basis when out with the child in public. Mostly this took a passive form with mothers describing how strangers frequently stared and whispered when they were out with their child. Occasionally it took a more active mode in the form of crass and insensitive remarks which were extremely painful for mothers, and to which they were particularly sensitive. Nor were such remarks confined to strangers and members of the public. One mother described how she would never forget an encounter with a hospital doctor in which he remarked

“well she won’t be your typical Irish dancer”.... [35: mother of child aged 4]

Many mothers described these situations and encounters as an inevitable aspect of life when you have a young child with a life-limiting neurodevelopmental disability. They were dealt with in different ways. Some mothers reported that they had learned to accept and become accustomed to these occurrences, while others expressed considerable anger and felt that they needed to confront the occurrences as they arose. It was hoped that confronting the situation or person would minimise the potential for recurrence

“like if you confront people then they won’t do it again....and if they *do* see a little special person then they won’t stare the way they stare at me...like that’s another one that won’t stare, do you know what I mean... it’s like you say to yourself “now *fuck you*”... [35: mother of child aged 4]

6.7.2.5 Interim Summary of Family Related Themes

These themes portray a picture of family life when a young child has a life-limiting neurodevelopmental disability. The child becomes the fulcrum around which all family life revolves, and the organisation of any family activity is dependent upon the child’s needs on any given day. Consequently mothers’ described significant impacts on all family members and all aspects of family life with family life is characterised by uncertainty and unpredictability.

The goal for mothers appeared to be bidirectional: mothers’ efforts were focused on keeping the child as well as possible, while simultaneously trying to maintain a sense of normality and minimise the impact on other family members. However negotiating a balance within the family, and a fit between the family and the community, was only a part of the picture for

these mothers who also needed to engage with and navigate the services that provided for the child's care.

6.7.3 Service Related Themes

All of the mothers who participated in these interviews had extensive experiences of both general paediatric services and disability services. However mothers appeared to differentiated between hospital based and community based services. There was the perception that the term "services" related to those services with which the child and family engaged regularly in their day-to-day lives. Although the children required frequent hospital admission mothers appeared to perceive this as quite separate to their "normal" services. While there were some differences amongst mothers in relation to their experiences of engagement with community based services, experiences of engagement with hospital based services were consistent amongst the mothers.

6.7.3.1 The Experience of Acute Services

Mothers described frequent episodes of hospitalisation of their child. Regardless of whether this was in a major paediatric hospital, a regional center, or a local tertiary hospital, these appeared to be a less than positive experience for mothers. Nor did the experience appear to improve over the course of the child's often numerous hospitalisations.

The major differentiating factor between admission of the child to a local hospital and admission to a regional or national center was the issue of expertise. Mothers believed that local hospitals often lacked the expertise and facilities necessary to provide the level of care that their child required. Two types of expertise were important: medical expertise and knowledge of local services. Medical expertise was essential to the appropriate management of the child's condition, and was highly valued by mothers especially if there was some consistency in terms of the hospital staff. Mothers described situations in which the difference in expertise, or medical opinion, between local hospitals and national centers resulted in difficulties for the child and the mother

"her medicine is another *big* thing....like every time she goes in they're altered...to [local hospital] like....but yet [national hospital] said that they are not to be messed around with.... that they put her on the *right* dose for her... just for instance the [drug] for her irritability to

calm her down...she was on that and I found it really good... but the doctor in [local hospital] reckoned that it was causing her to gag and wretch...she had this in her mind that this was the reason...and I said “it *couldn't* be because she is on it since she was a baby”....it couldn't be you know, cause [national hospital] hadn't changed the amount...but she had this in her head anyway so she stopped the drug and gave her a new one....but the new drug was administered under the tongue and [child] has an aversion to anything near her mouth....I mean that's why we ended up with the NG feeding and then the PEG feeding and everything like....so I was saying “*this is crazy*”...and the worst thing about it was she was getting her sleep medicine at about 9 o'clock or half nine and going to sleep... and I had to give her this new drug under her tongue at 11 o'clock while she was sleeping.... So I was nearly like going in pulling down the sheet and trying administer this medication.... and sure then she would be awake and hysterical again...it was nearly just you know....a *joke*....” [61: mother of child aged 2 ½]

It also appeared that health-professionals knowledge of the services that were available to the child and family at a local level was essential in order to refer the child and family to services that they would require on an ongoing basis. Where local services were organised from an acute hospital mothers described a much smoother and more seamless transition between services.

In all other respects hospital based services, regardless of where they were delivered, were described as sharing many of the same problems, with episodes of hospitalisation causing difficulties for mothers on a variety of levels. Although mothers described many practical difficulties associated with having a child in hospital (poor facilities, parking difficulties, increased costs, trying to organise other children at home) it was the process of engagement with hospital based services that appeared to be the greatest challenge for mothers. Several difficulties were described. These included the attitudes of medical staff, a lack of support for parents, poor communication (with the parent, with other disciplines in the hospital, and with the child's local services) and a general perception that the mother should fit with the needs of the hospital during the child's stay.

Mothers described attitudes of medical and nursing staff that were, at best paternalistic, and in many cases dismissive. All mothers described feeling that their unique knowledge of the child and their experience and expertise in the child's care was not valued or even

acknowledged by hospital services. This was particularly marked in non-specialist hospitals where mothers felt that they were probably more experienced in the management of the child's care than some of the medical staff they encountered. There was the perception amongst these mothers that failure to acknowledge their expertise could delay the commencement of appropriate management of the child's problem. One mother, whose daughter presented as she always did when her shunt became blocked, described the situation in a local hospital as follows

“and you know I was asking them to do a CT Scan.... she was having a major seizure.... like huge long seizure going on for over an hour because the ambulance couldn't find us.... and the guy just wasn't listening to me.... even though I tried, I could *see* he was not listening to me.... and it took *three days* in there before she actually had a scan which showed immediately that her shunt was blocked... so that was *really upsetting* and I wrote letters and stuff like that but.... [25: mother of child aged 6]

Mothers also described what they believed to be an expectation by the nursing staff on the ward that the mother should make up for deficiencies in the service and facilitate what mothers believed was insufficient staffing by providing much of the hands-on care and observation that the child required during the hospitalisation. There appeared to be relatively little acknowledgement that mothers may have obligations outside the hospital environment. A mother, who had three other young children at home, described this situation

“I used to go home for a few hours in the evening... for the other kids.....they [nurses] would say to me that I went home at an awkward hour...I would go home between six and nine, and they said that they had a changeover between eight and eight thirty....” [04: mother of child aged 4 ½]

Poor communication was a major issue described by mothers in relation to hospital services. Communication at every level appeared to cause difficulties. Mothers felt that communication with parents was lacking, and a lack of communication between different health-professionals in the hospital was a cause of several difficulties. More than one mother described how planned surgery for the child was cancelled on the morning of operation because of miscommunication between members of the hospital team. In addition this, failure of different hospital based teams to communicate about the child meant that mothers

constantly had to repeat the child's history from beginning to end. This was a cause of considerable frustration for some mothers

“and then....after *all* of that, the anesthetist came down and asked had [child] been well..... and I was like “*what the fuck*” would you read the filecan you not see the *size* of it like.....[16: mother of child aged 3]

The difficulties of poor communication were pervasive throughout mothers' stories, and were not confined only to episodes of hospitalisation. Mothers described difficulties associated with failure of communication between hospitals, and between hospitals and primary care services. Two mothers proposed solutions to this problem. Both suggested that this particular problem could be ameliorated if parents held an official record of their child's care.

Given the difficulties associated with episodes of hospitalisation it is probably unsurprising that the longer the child was hospitalised the more difficult it appeared to be for the mother to carry on in that environment

“but at that point we had been there 4 weeks and I was just absolutely *broken*... that's is the only word I can put on it.... I just came to breaking point....I just said “I can't stay here anymore... I *can't* do it.....” [39: mother of child aged 4 ½]

Despite the difficulties encountered during hospital stays mothers did not describe any difficulties associated with accessing acute hospital based services during a crisis in the child's condition as this usually meant taking the child to an Accident and Emergency department [A&E]. Because the child was ill they were generally not subjected to long delays waiting to be seen in the A&E. This was not the situation with regards to obtaining appointments and investigations however, where mothers reported long delays and difficulties in respect to this aspect of hospital based services.

6.7.3.2 The Experience of Community Based Services

Experiences of community based services were not consistent across all of the interviews. Three mothers described that they were “very happy” with the community based services they received⁹. These families lived in different parts of the country and the child's diagnoses

⁹ These included mothers of children aged 2, 3 and 6. Geographical area included Munster and Leinster. However, all of these mothers had services that were local, had been organised through the child's doctor, and all had a named professional person they could call on to organise additional requirements if needed.

and ages were not the same. Geographical variations in the services provided may have been responsible for this situation. Several instances of geographical inequality were described by mothers, which in many cases were a cause of resentment

“if we were living in the [next county] we’d have [service] now... and I have contacted [service] *several* times to see if they can’t help in any way.... but because they are not in this area they can’t.....no..... none whatsoever..... [61: mother of child aged 2 ½]

Mothers knew that neither the quantity nor quality of services were uniform across the country from speaking with other parents who, although in situations similar to their own, received different services. The remaining interviews were consistent in their description of community based services. All of the remaining mothers expressed a desire for better, more frequent services that were easier to access, less bureaucratic and more attuned to the child’s individual needs.

Mothers described community based services that were both insufficient and inconsistent. All mothers spoke of the “battle” to get the services they believed their child required. This metaphor consistently arose in interviews relative to mothers’ attempts to access community based services (although the metaphor originally arose in the surveys in Stage One). There appeared to be a marked disparity between what the child and family needed and what the services could provide. Although mothers described that more services of all kinds were needed this was particularly marked in relation to ongoing therapeutic interventions for the child, most notably physiotherapy and occupational therapy

“if she needs more physiotherapy why can’t she get it?....if it’s going to help *her* in the long run *and* make things easier on me why doesn’t she get it.... I don’t understand it.... [61: mother of child aged 2 ½]

Mostly mothers attributed this lack of essential services to cutbacks, funding shortfalls, and a moratorium on staff recruitment. They suggested that not only was there insufficient funding to operate services, but there were insufficient health professionals to deliver such services. Therapists were not replaced when on maternity leave, annual leave, sick leave etc. and so no service was available to the child during these occasions. In addition some mothers expressed the belief that their child’s limited potential for improving caused them to be relegated to

second place when scarce services were being rationed. This could take the form of services not being offered to the child

“for me I feel that the mindset is that the more profound the child is.... the less they have to do... I suppose dealing with the mind set of people that you know when your child is *so* profoundly disabled that mindset that well there is not a lot we can do here for this child, so basically lets just give them the bare minimum” [34: mother of child aged 2]

or the form of services being withdrawn from the child

“and for a long time I fought to get [child] Hydro Therapy..... later there was the reasoning that because [child] needs one to one care and there is not always a nurse that can take him down.... my argument.... I had a *lot* of tears a lot of *battles* over this... but my whole argument was you know you are a moderate to severe service and you know later on [child] will still be hereand all those other kids you have taken down to Hydro Therapy now... I said they’ll all be gone... I can understand you know we are all special needs parents... but you know these children will all be going on to other services and will have some kind of life outside.... and I said *here* is a child who is severe..... who will be *remaining* in your services.... *how* can you turn around at this stage and say to him he is not offered what every other child is offered.... I said there is one thing that he really, really enjoys...why is it *every* other child here gets it apart from mine.[04: mother of child aged 4 ½]

In addition to inconsistent services, mothers also described the difficulties they encountered with transient and inconsistent staff. This was perceived as a problem for both the child and the mother. Mothers found it easier to trust individuals who they felt knew the child. A trusting relationship developed as the person worked with the child and family over a period of time. Many mothers described how this was very difficult in terms of high staff turnover and the perception that “it is only a job” to people who were only intermittently involved with the child’s care. Mothers also described how they believed staff consistency had a positive impact upon the care the child receives.

Along with the services required by the child, mothers described services that they needed to support them in their caring role, but which were also lacking. Many mothers felt that in-home support services were insufficient, inflexible to the needs of the child and family, and in some cases were available in name only. Mothers described how support services seemed to lack any proactive focus, appearing to be orientated towards crisis intervention only

“I think “why let things go so bad that you’re down and out and you’re depressed”.....why wait till *then*.....for a crisis to happen... or be on the *verge* of one....they *know* [child] is very special, they *know* how difficult she is and how hard she is.... she is 24 hour care... but yet throw in an hour in the day time...It’s an insult..... [61: mother of child aged 2 ½]

Mothers outlined how the in-home support hours allocated to the family were insufficient to meet their needs. They described having little or no choice in relation to either the number of hours provided, or the manner in which the hours were delivered. It also appeared that home support services could be reduced or withdrawn from families without justification or explanation. Withdrawal, or the constant threat of withdrawal, of in-home services was a source of constant anxiety for mothers

“[service] is gone now this month, this is me last month to get itso that’s when [service] ends and [home support worker] is gone.... I have *no* help now... my help is gone.....and I do be panicky... thinking like I am on my own now, now I’m *really* on my own... [35: mother of child aged 3]

Many described being so desperate for any kind of in-home support that they could not distinguish between good and bad services.

Even a small reduction in home support hours was significant as it appeared to increase the burden on the mother exponentially. One mother described how, when she questioned the decision to reduce her support hours, she was made to feel guilty and selfish

“I need home support, and not being made to feel *guilty* about it..... or not feeling that I don’t *really* deserve it because my child is not that bad.....I suppose like to be honest the answer I have had is that I am bottom priority at the moment..... and I *am*, I mean there are people out there who are really, really having a tough time...I *know* that.... but it doesn’t mean that *I’m* not....” (25: mother of child aged 6]

There also appeared to some confusion relative to the demarcation of some in-home support roles. This was particularly problematic when one individual was performing more than one role (for example home support worker on one day and home help on another). Mothers reported that it could be difficult to be exact about the remit of the role, and the adherence to rigid, and in some cases nonsensical, rules about what jobs could be performed were a cause of frustration

“I think they don’t really know their job..... there is so much *confusion*.... they are not allowed to this, they are not allowed to do that.... they *are* allowed clean out the fire, but they’re *not* allowed to clean out the fridge.... It’s....*crazy*....” [61: mother of child aged 2 ½]

Respite services were another complex issue for mothers. Only four families availed of out-of-home respite care for their child. For these families respite was absolutely essential to continued coping, although all of these mothers still felt the need to explain, or justify, the decision to use it. Out-of-home respite was not always an uncomplicated affair. While one mother described respite that was easy to access, flexible, and available whenever it was needed, another described respite that was almost impossible to avail of, extremely rigid, and so difficult to access that it was hardly worth while

“I have respite in the [service] in [city]¹⁰..... so I have to drive to [city]in [local town] I have one weekend a year... well I haven’t even got it, the *application* is gone in for it, so..... but its only one weekend a year.....”

Interviewer: And how often have you got [city]?

“I haven’t had..... we couldn’t use it..... last May was the last time we used it.... because if [child] has a head cold or flu or anything she is not allowed to go..... she has to be well, and she hasn’t *been* well.... she has been in and out of hospital..... and if [sibling] had been sick if she had a bug or something you can’t just bring [child] up because there are sicker children there.....So we haven’t been able to.... use it since last May..... May 09 [61: mother of child aged 2 ½]

One mother, who had two children with a congenital condition that caused them to almost never sleep, described how respite had recently been offered for both children, but on different weekends. This was of little practical value to the family, and they were waiting to see if their request for a shared weekend could be accommodated.

In-home respite was no less complicated as the complex care required by the child made getting a suitable carer very difficult. Often a qualified professional carer was required but their availability was limited. This resulted in in-home respite hours being organised to suit the needs of the carer rather than the needs of the family

¹⁰ Travelling to this respite facility involved a 149 mile round trip for the mother.

“at the minute I have one night which you see.... then because she is a *nurse* and she is a pediatric nurse in [local hospital] .. you know let’s say I have a really *bad* two nights and I want her tomorrow night and she is working in the hospital..... then I *can’t* get her ...so then the days that..... *she* picks the days to come.....sometimes it suits and other times it doesn’t you know..... the *majority* of times now it wouldn’t suit..... like she was on holidays two weeks last week or the week before... but when she came back [child] and [sibling] had a bug.... and she couldn’t come then *either* cause like she has two kids of her own.....what can you do...” [61: mother of child aged 2 ½]

Overall community based services appeared to have two expectations of mothers: firstly that they should make up deficiencies and shortfalls in services, and secondly that they should fit with the service that was provided rather than providing a service that fitted the needs of the child and family. Where services were absent or insufficient mothers were expected to make up this deficit

“So they were trying to employ one [occupational therapist] and they did get one on board..... I think it was about 3 months afterwards an OT came on board..... and then she got pregnant so she left, and they never replaced her, so for 9 months then.... after only having an physio for about 4 or 5 months, for 9 months there was none.....they just kept telling me “you *got* the programme off [therapist] can you not work with that?....” [34: mother of child aged 2]

Many mothers reported a “take it or leave it” attitude from services. They also described incidences of obstinate inflexibility which appeared to demonstrate little understanding or empathy for the child and mother’s situation

“it was my kid’s sports day and I dropped him 12 minutes earlier..... and I was told “your time is allocated at 9.30” and it was 18 minutes past..... and they said “if you want to come in any earlier you are going to have to ok it with the Supervisor”..... and I said “look it’s because usually [child] sleeps in the car” and I *explained* that he didn’t sleep today and I didn’t want him to be just sitting in his chair..... and I thought I would just bring him in, and I don’t have to *leave* until 9.30..... I can stay with him it’s just he *wanted* to come, he recognized the door he was trying to pull at the door to go in... I didn’t want to.... if I tried to take him way he would have fought against it..... he would have just started to lie on the floor and I would be upset..... so the easiest thing was..... but they were *very, very* hesitant to open the door ... [04: mother of child aged 4 ½]

Mothers also depicted significant problems accessing the equipment the child required to support them in their day to day lives. Obtaining necessary equipment involved considerable effort and substantial waiting periods on the part of the mother

“we had to wait an *obscene* amount of time to get it....it was *months and months and months....* of ringing and ringing and ringing...” [38: mother of child aged 3]

In many cases the process of obtaining equipment was so protracted that the equipment no longer met the child’s needs by the time it was available, while in others cases suitable equipment was simply not available due to insufficient funding. Frequent encounters with bureaucratic wrangling and cumbersome processes were also portrayed. These made life incredibly difficult and appeared to be a significant source of stress. One mother proclaimed

“Like it’s the *system* that would have an awful lot to answer for in the stress in your life.....do you know what I mean, *that* would be the cause of the majority of the stress.....” [35: mother of child aged 4]

Many mothers explained how bureaucracy contributed to their overall burden of care. They described the difficulty of constantly having to reapply for the child’s entitlements despite the fact that it was obvious that the child’s circumstances were not going to change, and they described being shunted from department to department when trying to access necessary equipment and services

“and you have to keep ringing up and ringing up and ringing up.... like I had to get a house phone for the simple reason that to make these phone calls that I make..... because they keep you on hold for so long.... they would say “you are through to the wrong department you have to go through such-and-such department to get that” ... and you have to deal with somebody else then for the hoists and then for the equipment you have to go through *another* department ... *another* one for your tubes..... and you say to yourself *I’m* the one with the sick child.... they just make life harder for people...” [35: mother of girl aged 4]

Paper-work was an additional cause of stress and frustration. Mothers described a process that was not only difficult and time consuming of itself, but often ineffectual and frustrating as completed forms were misplaced or went missing and the process needed to be repeated or resulted in long delays

“Oh it’s *terrible* do you know..... it is... just like I was sick for a *week* trying to fill up that form... so then this letter thing came in the post and when I saw it I just thought “Jesus why

did I *bother*.... why didn't I just leave it, suffer on"..... its just all *forms*..... its all *pressure*.... its all "you have to have a form" for *everything*.... [45: mother of child aged 4]

At a general level there was a perception amongst some mothers that general disability services are not structured to accommodate the type and level of complex medical care that their child required. This was particularly evident in cases where the child was of an age to be involved with education services. Mothers described what appeared to be a conflict between two philosophies of care which resulted in the holistic needs of the child being overlooked particularly where the child had multiple and complex needs. This could also result in poor management of the child's symptoms and an additional burden on the mother

"I had to fight and fight to get her back into preschool because her preschool has never had such a complex child..... they never *had* a child on Morphine there... they wouldn't have as *sick* as [child]... cause even though she is special needs she is a *sick* special needs child...so like there is a control drugs book do you know ...when you hand in Morphine and stuff.... and they have never had that so they still don't have it in the school because they have never had a child on Morphine..... so it was meeting after meeting after meetings.... but they still don't have the control drugs book so if [child] gets a pain now *I* have to go to the school and give her the medicines... because they do not have this drug book and they are not able to give the drugs in the school.....so I am kinda putting her in to school and then you have to always have to have your phone knowing that she could need Morphine or she could need Oromorph..... now they could give her Paralink..... but then needing Morphine and getting Paralink.... I mean that doesn't...there's no *sense*....." [35: mother of child aged 4]

6.7.3.3 General Service Related Issues

The previous two themes describe mothers' experiences of hospital and community based services. These were presented separately because this was the manner in which mothers perceived and described them. However there were also issues that crossed this border of distinction and related more globally to issues of service provision. These included difficulties making contact with services, and the importance of individuals within services.

Mothers reported significant difficulties they experienced in relation to making contact with services, both hospital and community based. Telephone communication in particular appeared to be a cause of considerable frustration. Mothers described many instances of

telephones persistently ringing out, excessively long periods of being left “on-hold”, not knowing who they should be asking to speak to or the person never being available, being transferred from department to department, constantly being “cut-off” and leaving multiple messages on answer machines but never receiving return calls. Not only did mothers feel that this was unnecessary and wasteful of their time, but many reported feeling angry, frustrated and humiliated. This is clearly evident in the following description

“phone calls and phone calls, and phone calls..... *Jesus*..... when I finally got through I said “hey can I speak to such and such a person” and they say “who is calling” and I’d say “[mum’s name]” ... of course then you get “sorry she is out of the office at the moment”.....I mean it’s very *convenient*.... so I would ring back in five minutes later and say “hey its Maria O’Neill or somebody” she would come to *that* phone..... so they start to avoid you do you know what I mean..... so then I started “how come you were not in a minute ago” and you *know* that they are lying and then I loose the rag.... I mean you do.... you have to make a tinker out of yourself..... you do.... You *have* to get in to these little fights with people..... but they are being paid for that, I am not being paid to be putting up with “hold on”, “hold on”, “hold the line”, “*fuck off*” do you know what I mean, you don’t have *time* to be sitting all day on phones to be making an ejit of yourself..... and you don’t want to be *lowering* yourself as a person to these people either.... [35: mother of child aged 4]

The second issue that mothers reported which related globally to the services the family received was much more positive. This related to the distinction between the “service” and some of the “people” who delivered it. Just as mothers could describe specific incidences in which strangers had been kind and helpful, all mothers similarly reported individuals who they had encountered in services who were flexible, caring and sympathetic, and who worked outside the remit of their role to support the family. These individuals with their positive attitudes and orientations came from all fields of services and were highly valued and appreciated by mothers. One mother described the profound impact that such individuals have on the child and family

“it’s just like sometimes there’s an *extra* person to love him for all that he is....it will alleviate some of that.... it’s like it’s *not* just me and his dad that loves him..... it is somebody else.....and this carer is so *good*.....it’s funny the way people affect your life..... [04: mother of child aged 4 ½]

6.7.3.4 Interim Summary of Service Related Themes

These themes present mothers' experiences of engagement with the services involved in the care of the child and family. Mothers' experiences of hospital based services were consistent, and unfortunately generally not positive. Mothers described a lack of specialist expertise at a local level, poor facilities for parents of hospitalised children, paternalistic attitudes from staff, a lack of a partnership orientation, and poor communication at all levels.

Alternatively mothers' descriptions of their experiences with community based services were inconsistent. While a minority of mothers were happy with the services they received the majority described services that are insufficient, inconsistent, bureaucratic and inflexible to the needs of the child and family. Despite this, all mothers identified exceptional individuals within services who worked hard, often outside the remit of their role, to make a positive impact on the child and family's situation.

6.7.4 Super Ordinate Themes

The themes related to focal areas of the study have been described. These have helped to explain and expand the findings from Phase One. However, consistent across, and overarching, the entire data corpus, two super ordinate themes were obvious in mothers' stories. The first of these, "Waging a War", describes the myriad of battles that families of young children with life-limiting neurodevelopmental disabilities encounter on an almost daily basis. The second, "Just Getting on With It", describes families' experiences of the management of everyday family life, and their experiences of engagement with services. These super ordinate themes help explain in a more three-dimensional way the experiences of the families who participated in this stage of the study.

6.7.4.1 Waging a War

"I have learned along the way that you *do* have to fight....and *not feel bad* about fighting....there's been a *lot* of tears.....a *lot* of battles..... and some days you're *able* to fight the fight, and some days you just can't..." [04: mother of child aged 4 ½]

This theme relates to the ongoing battles faced by families of young children with life-limiting neurodevelopmental disabilities. The theme was consistent across every mother's

story. Battles large and small were fought on an almost daily basis; they were motivated by the best interests of the child, and fought to obtain what the child needed. These battles took place on many levels. Mothers battled the instability and uncertainty of the child's condition; they battled to reconcile the needs of all members of the family. Families battled to maintain a sense of normality and stay afloat as a family while managing the many demands of day-to-day care; they battled with services to get what their child needed; they battled the bureaucracy of services that made life unnecessarily difficult; and they battled the social prejudices, perceptions and difficulties they often encountered. Individual battles were won and lost but the long-term war continued. Winning a battle achieved something positive for the child: stabilization of the child's health condition, a better service, equipment that was needed, confronting a hurtful or crass comment or remark, while losing a battle meant that the child was overlooked or forgotten. Mothers were anxious to emphasise that this war was absolutely necessary. It was fought for, and motivated by, only the child's needs and entitlements. This was a position that they would not have chosen if it had not been forced upon them. Fighting was something they had been required to learn as a result of their experiences with their child

“always pushing I suppose..... you learn you *have* to push.... the person who asks it the person who gets.....it doesn't just come.....and if you *don't* ask then you *don't* get... you know maybe you might like to think that they see my child needs this and this... and may be they *will* just offer it to me and I *won't* have to go and say “why aren't you giving this to my child”.... but you *do*.... You *have* to fight, you *have* to ask..... you have to demand “why not my child, my child needs things too.....” [04: mother of child aged 4 ½]

As in any conflict strategic planning was important. There were occasions in which mothers described having to “pick your battles”. Limited resources and lack of funding meant that it was not always possible to get exactly what was needed and in these cases mothers prioritised, and fought for, what was most important to the child at that particular time.

There were allies who could be called upon when help was needed. For the most part family and friends could be galvanized for psychological and practical support. Other parents, with experiences similar to the mothers in this study, provided much valued information and advice on how to navigate the many services involved in the care of the child and how best to obtain what was needed. There were always exceptional individuals in services who could be relied upon to ameliorate the difficulties.

Mothers stressed that the battle was against “the system” rather than against any particular service or individual. They emphasised that they did not want this constant state of conflict, however necessary, to define them and hoped that their adversaries could distinguish between the mother fighting for her child and family, and the person who was that mother

“I don’t want to be seen as that parent....coming in and constantly demanding all these things... I don’t want to be *that*... I’m *not* that person really...”[04: mother of child aged 4 ½]

6.7.4.2 Getting On With It

This theme relates to families experience of everyday life when a young child has a life-limited neurodevelopmental disability, and to the main orientation of services towards the family. Mothers narratives described how eventually there came a time when they had little choice but to roll up their sleeves and get on with the task in hand

“eventually it’s like “right, I *have* to do this now ”.....and then it just comesI don’t know *how* but it does.... you just *get up* and *get on* with it” [12: mother of child aged 1]

Mothers’ stories suggested that they “got on with it” out of a combination of love and devotion, a sense of duty, and a lack of choice. Fortified with technical skill, increased confidence, a sense of personal strength, and a focus on the positive elements of the situation mothers managed to keep their child and family going day by day. It was also evident from some mothers’ stories that “getting on with it” was also what services expected them to do

“it came to the stage that she was going in and out of hospital *so* much because she was *so* irritable and *so* upset that they would say “you can’t keep coming in her all the time now”.... they thought that was just the way [child] was going to be... [61: mother of child aged 2 ½]

While good services provided the resources that empowered mothers to do this, often insufficient, inconsistent and bureaucratic services left them with no alternative

“but it gets to the stage where you just say.... *not* that anybody listens...you now when you are fighting, fighting, fighting nobody *listens*..... so you just get up and get on with it do you know what I mean..... because *nobody* is going to say “well hold on I will listen to you” do you know what I mean... nobody is going to say “o.k ... tell *me* what is your problem”.... because nobody does... [35: mother of child aged 4]

This expectation that mothers get on with it was rarely explicitly stated. The conveyance of the message was generally subtle, and often cloaked in platitudes and rhetoric of concern for the mother

“and between the *team* and that *liaison nurse* I was talking about and the girls from [*service*] they would all be saying “now *promise* me that you will look after yourself”... you know and “do this, and do that, and the other”....but no one was willing to give me extra *help* all the same....” [61: mother of girl aged 2 ½]

Getting on with it is exactly what mothers in this study did. They got on with the complicated task of caring for their child; they loved the child for who they were while accepting the reality of their circumstances; they got on with maintaining a sense of normality for other children and for the family itself; they got on with the task of making up for deficiencies in services while continuing to battle for better services for their child. Mothers in this study got on with doing the very best they could, and remarkably many even managed to have sufficient energy left over from coping with the demands of their own lives to provide support to other families in situations similar to their own.

6.8 Summary of Key Findings from Phase One, Stage Two

The findings from this stage of the study suggest that young children with life-limiting neurodevelopmental disabilities suffer considerable physical and social morbidity as a consequence of their condition. The child’s condition requires mothers to provide complex medical and technological care, often on a 24-hour basis, and for which they receive little training. Mothers describe being overwhelmed and overburdened, particularly in the early stages of coming home from hospital with their child, when community services have yet to be mobilized and there is little support for the family from local health services.

Family life was characterised by unpredictability and uncertainty, with families struggling to maintain as normal a life as possible for all family members. In order to overcome the many practical challenges that are encountered on a daily basis family life was planned and executed with military precision. Internal and external resources were mobilized, and partners, friends, family, and other parents in similar situations were called upon to provide

practical assistance, information and support; although it would appear that the mothers assume the main role of care coordinator.

Oscillations in the child's condition resulted in numerous episodes of hospitalisation, and were a cause of suffering for both the child and the mother. The ongoing care of the child had negative consequences for other family members. Mothers suffered physically, psychologically and socially as a consequence of the provision of care. Siblings suffered the loss of opportunities for normal family interaction. Relationships between parents were also impacted. In addition, the family unit experienced financial strain and the challenges of losing their home environment as it was consumed by equipment needed for the care of the child. Outside the context of the family families inhabited an outside world that appeared insensitive and lacking in understanding.

Experiences of hospital based services were consistent, and were generally not positive. Mothers described poor facilities, paternalistic attitudes, lack of partnership, and poor communication. Experiences of community based services were inconsistent. While a minority of mothers were happy with the services they received the majority reported services that are insufficient, inconsistent, bureaucratic and inflexible. The lack of available, consistent, dependable services was a constant and significant source of anxiety for mothers.

The overall experience of caring for a young child with a life-limiting disability is characterised by a constant struggle. Mothers managed on an ongoing basis because there was no alternative. Yet despite all of the hardships and challenges encountered mothers focused on the love and devotion they gave to, and received from, their child, and on the positive elements of the family situation.

6.9 Preliminary Discussion

Without exception mothers' accounts of providing care to their children with life-limiting neurodevelopmental disability are poignant. The themes and super ordinate themes inherent in mothers' narratives in this study are resonant with other qualitative work in the area of childhood disability and childhood life-limiting illness, and the findings are generally

consistent with other published research in this area (O'Brien, 2001, Alexander *et al*, 2002, Redmond & Richardson, 2003, Steele & Davies, 2006).

The difficulties and challenges experienced by the children in the sample for this study have been enumerated in Chapter Five. This quantification however does not necessarily provide an insight into either the complexity of these children's conditions, or the associated level of care they require. Nor does it provide an insight into how the children are valued, cherished, and incorporated into the family unit. These issues arose as important features of the interview data. The burden of care for a disabled child with complex needs is widely reported in published research literature (Redmond & Richardson, 2003; Quinn *et al*, 2005; Erickson-Warfield & Gulley, 2006; Monterosso *et al*, 2007). Research shows that assessment of the child's general state of health, medical treatments, and time-consuming care activities are common and integrated parts of the child's care (Kirk, 2001; Nicholl, 2008; Ouellet, 2009) which result in financial burden, burden due to effects on family routine, and burden due to the effects on the physical and mental health of other family members (Datta *et al*, 2002, Steele & Davies, 2006). The provision of high levels of skilled care and expertise by mothers of chronically ill children who require repeated hospitalisation has also been previously documented (Balling & McCubbin, 2001) and similar findings have been reported in the context of complex disability by Redmond and Richardson (2003), children with life-limiting illnesses (Quinn *et al*, 2005) and children with complex healthcare needs (Nicholl, 2008).

Mothers descriptions of the exceptionality of their child echoes previous research related to parents' positive perceptions of a child with a disability, and their descriptions of caring for their child as both a source of distress and enrichment is consistent with previous research findings (Kearny & Griffin, 2001, Carnevale, *et al*, 2006; Trute *et al*, 2010). These findings illustrate the conflicting, and usually concurrent emotions, mothers' experience, whereby they report both positive psychological states (joy, love, commitment, pride, hope) and negative psychological states (anxiety, frustration, distress) as simultaneous aspects of care giving. Some of the positive themes previously identified are recognisable in the narratives of mothers in this study, especially those descriptions of the child as a source of joy and happiness; increased sense of purpose; a source of personal growth and strength; and a source of increased tolerance and understanding; and expanded personal networks (Stainton & Besser, 1998; Green, 2007a). In a study of medically fragile children Patterson and Garwick

(1994) suggests that mothers selectively attend to the positive aspects of their child's personality and behaviours while minimizing the limitations of the health problems. Minimising the health problems of their children was not evident in mothers' narratives in this study, possibly because the complexity of the child's condition and the associated care required meant that this was not an option for these mothers. However, consistent with Patterson and Garwick (1994) mothers did focus on the warmth and responsiveness of the child, the child's tenacity and perseverance, and the positive ways in which the child impacted on other family members.

Burr and Klein (1994) describe a conceptual framework of coping strategies that families use to manage adverse events and family stresses, many of which are evident in mothers' descriptions of their experiences of caring for their child. Mothers' demonstrated the use of cognitive coping skills in their acceptance of their situation, attempts to gain useful knowledge, and their increased independence and self-sufficiency. They demonstrated emotional coping skills in their expressed feeling and affection for their child, and focus on the positive aspects and elements of their situation. They demonstrated relationship coping in their renegotiating of their relationships with their partners and increased adaptability. Many of the coping approaches to the ongoing management of family life that mothers describe in this stage of the study are similar to those described in the stress and coping literature (Affleck *et al*, 1993; Turnbull *et al*, 1993; Park & Folkman, 1997; Fredrickson, 2001). Folkman's (1997) process of meaning making was evident in mothers' narratives of positive reappraisal and infusing events with positive meaning. Their descriptions of these processes supports Hauskov-Graungaard *et al*'s (2011) model of resource creation, whereby that the stressors involved in caring for a child with severe disability are so pervasive and ambiguous, and challenge main beliefs and goals to such an extent, that resource creation becomes a basic necessity for coping with the situation. The concept of "giving something back" was important to mothers in this study, and although Nicholl (2008) also reports this phenomena in a study of mothers of children with complex needs she suggests that the mothers in her study felt under some "real or perceived pressure to do some fundraising for the organisations or services that provided help or services" (p.213) and that this added a new dimension to the phenomena of care-giving. This was not the motivating factor for mothers in this study.

The process of normalization as a coping mechanism has been reported in research literature related to children with severe disabilities and technology-dependency (Carnevale *et al*, 2006; Hauskov-Graungaard *et al*, 2011). Mothers in this study reinforced the concept of their family as normal through peer-support. Peer-support has been identified as an important coping resource for parents of disabled children (Case, 2000) and for parents of children with complex needs (Carter *et al*, 2007), although generally this has been reported in the context of information gathering and providing social opportunities for parents.

The general strategies that mothers used to manage day to day are similar to those identified by Clarke-Steffen (1997) in a study of mothers of children diagnosed with cancer, particularly in relation to reorganizing roles, evaluating and shifting priorities, assigning meaning to the illness, and managing therapeutic regimens. Mothers descriptions of day to day family management exemplify the Enduring family management style described by Knafl *et al* (1996) and Knafl and Detrick (2003) in which families of a child with a chronic condition describe themselves as burdened but confident of their ability to manage whilst at the same time focused on the tremendous effort required to adhere to the treatment regime. There is a sense in which the experience of life within the family, trying to manage the situation but hoping that things will get better, mirrors the experience of the interaction between the family and the outside world of services.

There are also parallels between this study's subtheme "Keeping the Show on the Road" and Seele and Davies's (2006) theme "Holding the Fort". Steele and Davis describe a grounded theory study of the experiences of families caring for a child with a progressive, life-threatening illness that are characterised by providing physical care, living by the clock, and coordinating services. Mothers' narratives in this study also resonate with O'Brien's (2001) descriptions of the dimensions of family life with long-term childhood technology dependence which she categorises into four dimensions: making sense of life; managing daily life; maintaining a functioning family; and negotiating with outside entities. The concept of battling and struggling has also been previously reported in the literature and is consistent with mothers' narratives in this study. Alexander *et al* (2002) describe "struggling daily" as a major theme to emerge from their grounded theory study of mothers of children with complex needs.

Mothers' descriptions of their experiences with community services are generally consistent with research in this area (Redmond & Richardson, 2003; Hunt, Elston & Galloway, 2003; Quinn *et al*, 2005; Ouellet, 2009) particularly in relation to difficulties associated with communication, approach to care, accessing essential services and equipment and bureaucracy. Watson *et al* (2002) identifies inadequate discharge arrangements and poor liaison between acute and community health professionals amongst the barriers to effective multi-agency working in the care of children with complex health care needs. Both of these issues were evident in the findings of this study.

In contrast to Quinn *et al* (2005) mothers in this study did not report positive experiences of hospital based services. Mothers' concerns about what they perceived to be understaffing or unavailability of nurses to render care in hospital contexts are consistent with other reports of an unspoken assumption that parents would be available to assist in their child's care because nurses were often too busy (Warner, 2000; Balling & McCubbin, 2001; Avis & Reardon, 2008). The children in this study required repeated hospitalisations, the assumption by nurses that parents would be present and constantly available to deliver care at the bedside at all times placed an additional stress on mothers. This is consistent with previous research reporting that there is now an expectation that parents would remain with their child during hospitalisation and carry out the majority of the day-to-day care (Reeves, Timmons & Dampier 2006; Coyne, 2007; Coyne & Cowley, 2007).

6.10 Strengths and Limitations of Phase One, Stage Two

As with Stage One of the study, this stage has particular strengths and weaknesses that must be considered in the context of the conclusions that are drawn. Participants were drawn from the sample used in Stage One; the limitations of this sampling frame have already been discussed in Chapter Five. However, every effort was made to sample participants in an objective and representative way, and the procedure for selecting interviewees ensured that participants from all ranges of family impact scores were included in this stage of the study which facilitated as broad a perspective as possible.

The lack of gender representativeness may be considered a potential second limitation. This study presents mothers narratives, and is confined to a description of mothers' experiences of caring for their young child with a life-limited neurodevelopmental disability. It was not

intended to exclude fathers from this stage of the study. Although three fathers had returned consent forms indicating their willingness to participate in this stage, they were not selected in the process of random selection used to identify parents for interview. However, since previous research has indicated the mothers are more likely to be directly involved in the child's routine medical care (Green, 2007; Monterosso *et al*, 2007; Manaseri, 2008) there is considerable merit in capturing the experiences and opinions of mothers whose vital care work for their young children with life-limiting neurodevelopmental disabilities is often unacknowledged and undervalued.

6.11 Conclusion

This chapter has provided an overview of the qualitative stage of the first phase of this study. The research design, its implementation, and issues of quality have been discussed. The method of data analysis has been described.

The findings of this stage of the study expand and support the findings from the parents' survey data obtained in the previous stage, and completes the sequential explanatory design used in Phase One. The findings provide a more holistic picture of the experience of caring for a young child with a life-limiting neurodevelopmental disability, and the myriad ways in which this impacts on all members of the family and all dimensions of family life. Mothers' narratives in this stage are supportive of previous qualitative studies which have described mothers' experiences of caring for children with life-limiting conditions and complex disabilities.

Chapter 7: Phase Two: Delphi and Findings

“Experience is the oracle of truth; and where its responses are unequivocal they ought to be conclusive and sacred”

Alexander Hamilton & James Madison (1787)
The Federalist Papers No. 20, December 11

7.1 Introduction

This chapter presents an overview of Phase Two of the study and its findings. As previously indicated in Chapter Four this phase addresses questions about service providers’ perspectives on the nature and type of services provided to children with life-limiting neurodevelopmental disabilities and their families. It is concerned with obtaining expert opinion related to three broad topic areas and addresses the following research questions -

- What are the goals of care for children with life-limiting neurodevelopmental disabilities and their families?
- How well do current services meet the needs of these children and their families?
- What changes are necessary to current services to improve the care of children with life-limiting developmental disabilities and their families?

This phase utilises a Delphi design. Delphi is a consensus method which facilitates communication between and among a panel of experts, so that the process is effective and the group as a whole can deal with a complex problem (Linstone & Turoff, 1975; Fink *et al*, 1984; Mead & Moseley, 2001; Keeney *et al*, 2001; Vernon, 2009). Currently there is very limited evidence of expert opinion in this area to guide evidence-based decision making and the Delphi method is particularly suited to such situations (Hasson *et al*, 2000; Meade & Moseley, 2001; Powell, 2003; Cook, Brismee & Sizer, 2005). In addition, because the opinion of a nationally representative panel of experts was required, using Delphi eliminated the practical difficulties of trying to assemble a geographically dispersed group (Linstone & Turoff, 1975; Critcher & Gladstone, 1998), and the anonymity afforded by the method prevented any potential possibility that group dynamics and hierarchies might influence individuals to conform to group opinion (Beech, 1999; Mead & Moseley, 2001; Garavalia & Gredler, 2004).

The chapter is presented in two parts. The first provides both a theoretical and practical discussion of the issues that are considered critical to the implementation of Delphi, and applies these critical features to the current study. The second part of the chapter reports and discusses the findings of the Delphi. The findings will be integrated with those from Phase One of the study in the next chapter.

7.2 The Delphi Method

The Delphi method was developed for the purposes of military forecasting and planning in the early 1950s (Brown, Cochran & Dalkey, 1969). More recently Delphi has been used as a constructive method in facilitating controlled, rationale group communication to develop knowledge for decision making (du Plessis & Human, 2007). Ziglio (1996) proposes three situations in which Delphi is the method of choice (a) forecasting future trends, (b) investigating questions that have little or no historical information, and (c) addressing questions which require the consideration of numerous issues where there is a need for pooled judgment.

Multiple definitions of Delphi exist (McKenna, 1994a; Hasson *et al*, 2000; Mead & Moseley, 2001; Brink, 2002; Powell, 2003, 2004; De Villiers *et al*, 2005;) and Delphi has been variously labeled in the literature as a “technique”, a “process”, a “method”, an “exercise” and a “survey” (Crisp *et al*, 1999; Mullen, 2003). Linstone and Turoff (1975:3), while acknowledging the many disparate views as to which is the most appropriate or best procedure, provide a broad definition of Delphi as “a method for structuring a group communication process so the process is effective in allowing a group of individuals, as a whole, to deal with a complex problem”. Indeed so many variations of Delphi are used by researchers that McKenna (1994a) and Mead and Moseley (2001) propose the term Delphi “approach” as a more accurate description of its many practical applications. Research literature is replete with studies reporting the use of “modified” Delphi forms (Carnes, Mullinger & Underwood, 2010; Chang *et al* 2010; Sue Hoyt *et al*, 2010; Jeste *et al*, 2010; MacNeela *et al*, 2010, West, 2010; Wilson *et al*, 2010; Wilson & Moffett, 2010; Zeigler & Decker-Walters, 2010).

However, such modifications have been the focus of criticism as far back as the mid-1970s with several areas of concern identified. Sackman (1975) originally raised issues regarding the methodological rigor of modifications made to the original Delphi form, while Hasson *et al* (2000) suggest that the absence of universal guidelines for Delphi can result in methodological difficulties where judgments have to be made in the absence of sound guidance. Despite its increasing popularity in health research Keeney *et al* (2006) suggest that much confusion, disagreement and uncertainty exists concerning the basic principles intrinsic to the application of Delphi.

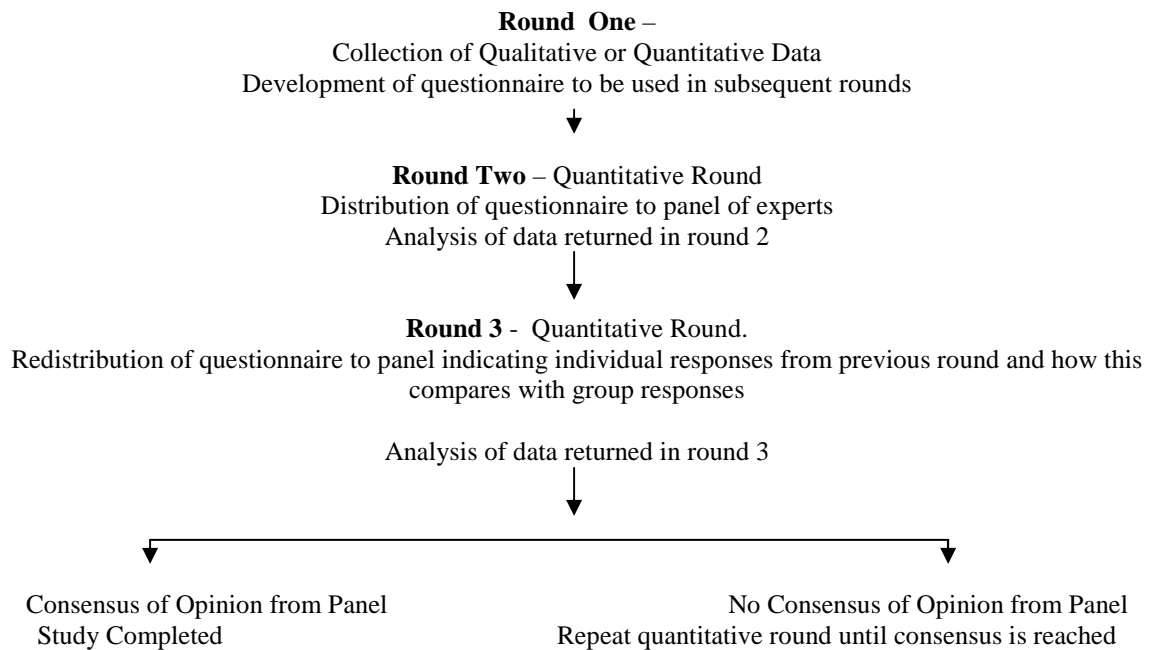
7.3 Core Features and Stages of a Delphi

Although widespread use of Delphi has resulted in various approaches, all applications describe common features of the technique, namely that it involves a series of sequential questionnaires or rounds, interspersed by controlled feedback, designed to elicit the most reliable consensus of opinion among a panel of experts. Dalkey (1969) identified the following essential features of the Delphi: anonymous response, whereby the opinion of group members are obtained by formal questionnaire; iteration and controlled feedback, whereby interaction is effected by a systematic exercise conducted in several iterations with carefully controlled feedback between rounds; and statistical group response, whereby the group opinion is defined as an appropriate aggregate of individual opinion on the final round. Du Plessis and Human (2007) add an additional essential characteristic - the selection of experts as respondents.

Mead and Moseley (2001:10) identify six stages involved in a conventional Delphi study similar to those originally identified by Turoff (1975). These include (1) selection of a panel of experts; (2) formulation of the question; (3) statement generation; (4) reduction and categorisation; (5) rating; and (6) analysis and iteration. In between the rating rounds a process of analysis of the received data takes place. The responses are collated and the original or revised questionnaire is re-circulated accompanied by an anonymised summary of responses (Mullen, 2003). The result is a “multistage” process whereby each iteration builds upon the results of the previous one (McKenna, 1994a; Keeney *et al*, 2001) [Figure 7.1]. Repeat rounds of the process are conducted with panelists invited to confirm or modify their previous responses in the context of the opinion of the group until consensus has been

reached or no further changes are taking place (Mullen, 2003). Consensus is assessed, or obtained, by asking respondents to accept, reject, rank or rate statements from the previous rounds (Stewart, 2001) thus the logic behind Delphi is partly statistical taking as a group response a statistical aggregate of the final answers (Dalkey, Brown & Cochran, 1969).

Figure 7.1 *The Delphi Process*



Adapted from du Plessis & Human (2007)

It is important to emphasize that the Delphi method differs from more traditional survey methodologies. While the descriptive function of much survey research is dependent upon instrumentation, measurement and observation, generally using sampling and inferential statistics to estimate the characteristics of a representative finite population from a random sample (De Vaus, 2002a; Bruce *et al*, 2008), in Delphi the desired goal is the securing of consensus on expert opinion. The following sections will consider individually each of the critical components of the Delphi process and discuss their application to the current study.

7.3.1 Selection of the Expert Panel

The credibility of the Delphi technique is heavily dependent upon the composition of the expert panel of respondents (Crisp *et al*, 1997; Hasson *et al*, 2000; Vernon, 2009), and a typical criticism leveled at the technique is the issue of how to choose a “good” respondent

group (Campbell *et al*, 1999). Critcher and Gladstone (1998) suggest that while the selection of the panel is essentially a subjective exercise, the key issues to be considered are the nature of each individual participant's interest, and the depth and range of expertise of the panel as a whole.

Although central to the credibility of Delphi studies, there is debate in the literature regarding the concept of "expertise" used in the Delphi approach. There is little consensus with regards to the definition of an expert, and inconsistency regarding the variable characteristics that an "expert" should possess (Fink *et al*, 1984; McKenna, 1994a; Green *et al*, 1999; Hasson *et al*, 2000). Even the term "expert" is open to interpretation and has been variously defined (Goodman, 1987; McKenna, 1994a; Green *et al*, 1999). Powell (2003) suggests that experts should be chosen for their work in the appropriate area and credibility with the target audience, while Goodman (1987) proposes that it would be more appropriate to recruit individuals who have knowledge of a particular topic and who are consequently willing to engage in discussion about it without the potentially misleading title of "expert". Rather than define the criteria for expertise Goodman (1987) suggests that the researcher has a responsibility to explicate and justify the selection procedures used to select participants and the basis on which they were chosen. Vernon (2009) concurs, suggesting that the criteria for expertise should be defined within the study context as it will vary depending upon the needs of the study. He suggests that the researcher's task is to define and justify the criteria used in their particular study.

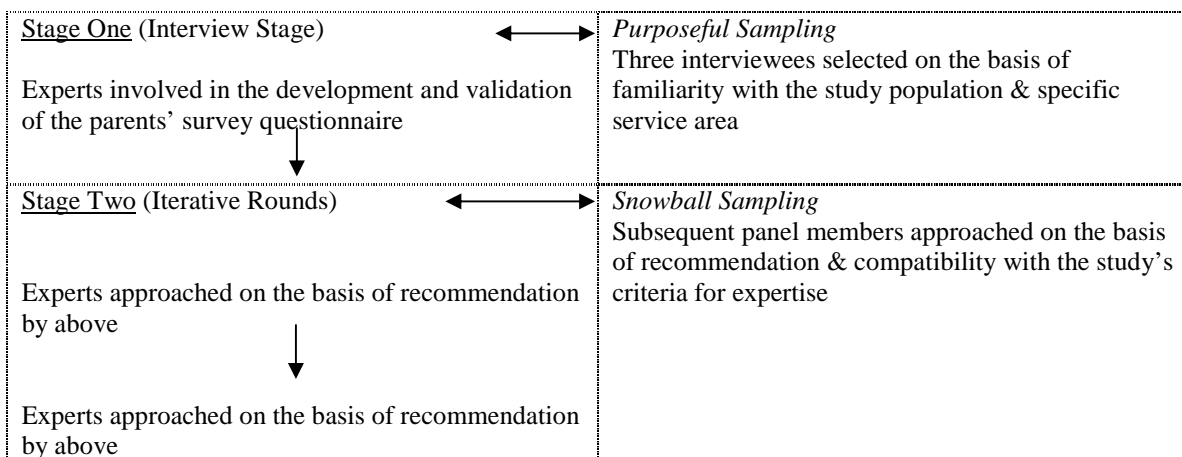
7.3.1.1 Selection of the Expert Panel for the Study

There were two elements to the selection of the expert panel for this study, with the sampling strategy involving a combination of purposeful and snowball sampling techniques [Figure 7.2]. The first involved the selection of three candidates to participate in qualitative interviews. These were chosen from the expert group who assisted with the development of the parents' questionnaire used in Phase One of the study. The purpose of conducting these interviews was twofold: firstly to obtain as much relevant and comprehensive information as possible in order to assist with the formulation of the questionnaire, and secondly to identify potential candidates for the formation of the expert panel who would participate in the iterative rounds of the questionnaire.

It was imperative that all services providing care to children with life-limiting neurodevelopmental disabilities and their families were represented in the interviews. These included three service groupings; general paediatric, intellectual disability and palliative care services, and the three expert interviewees were purposefully selected on the basis that each represented a different service group. This ensured a comprehensive and representative range of opinion was obtained during the interviews, and that a broad range of experts from a variety of services could be indentified for participation in the iterative Delphi rounds. Expert interviewees were not included in subsequent rounds of the Delphi questionnaire, although they did participate in the pilot of the various questionnaires and reviewed them for face and content validity.

The second element involved the identification of individuals to form the expert panel for this study. The group of experts for the iterative rounds of the Delphi were selected using a combination of purposeful and snowball sampling. Snowball sampling is a frequently used sampling strategy when the researcher does not have access to a population from which to draw a sample, or where the nature of the research project makes drawing a representative sample impossible (Dattalo, 2008; Nagy Hesse-Beiber & Leavy, 2010). The three interviewees were asked to identify other health professionals who they considered to be experts in the care of children with life-limiting neurodevelopmental disabilities and their families, and who met the study's inclusion criteria. Each individual identified was then asked to nominate other experts who also met the inclusion criteria for the study.

Figure 7.2 *Selection of the Expert Panel for the Study*



Where possible potential participants were initially contacted by telephone. This strategy was adopted in light of McKenna's (1994a, 1994b) and Vernon's (2009) suggestion that personal contact can increase recruitment and help reduce attrition rates in subsequent rounds.

However, given the nature of the respondent panel this was not always possible. It was difficult to make direct contact with some community based participants as they were often geographically mobile and without a stable base. In addition because the entire expert panel had a busy clinical remit contacting individuals at a convenient time was often extremely difficult. All potential participants received a letter inviting their involvement in the study [Appendix I] and an information leaflet outlining the details of the study. Included were details of the study's nature and objectives, the nature of the expert panel, the importance and obligations of participants, length of time the study would take, and what information would be shared amongst participants [Appendix J].

7.3.1.2 Criteria for Expertise in the Study

In this study various forms of expertise were needed and it was necessary to introduce as many relevant issues as possible into the study. Consequently input was solicited from a broad-based expert panel. Two key characteristics were sought in panel participants – Expertise and Credibility. In the context of expertise it was necessary to ensure that different points of view (e.g. in terms of discipline, practice experience etc.) relevant to the care of children with life-limiting neurodevelopmental disabilities and their families were adequately reflected so that informed and relevant opinion could be gathered. It was also essential that panelists were credible so that opinions would be taken seriously by others working in the area and beyond.

Credibility was demonstrated by the fact that individuals were recruited to the panel on the basis of having being identified as “expert” by a variety of their peers. This was reinforced as many panel members were recommended by more than one other expert in the field. The study considered an individual to be an “expert” if they met the following inclusion criteria –

- (1) Held a professionally recognised health-related qualification.
- (2) Had no less than five years experience providing care to children with life-limiting neurodevelopmental disabilities and their families.

7.3.2 Size of the Expert Panel

Once the nature of expertise required for the study has been defined, the sample size for each round of the Delphi needs to be decided. The sample size for constructing a Delphi panel is not a statistically bound decision (Atkins *et al*, 2005), and optimal panel size has not been established (Campbell *et al*, 1999; Powell, 2003; Vernon, 2009). Consequently it is clear from the research literature that there are wide variations in the numbers used. Both Dalkey *et al* (1969) and Linstone (1978) propose seven as a suitable panel size suggesting accuracy deteriorates rapidly with smaller sizes and improves more slowly with larger numbers. Turoff (1975) recommends a minimum of 10 and a maximum of 50 on the panel. Delbecq *et al* (1975) advises that with a homogeneous group of people 10 to 15 participants may be enough. Ziglio (1996) concurs, suggesting that good results can be obtained with a relatively small group of homogenous experts, although without specifying what number constitutes a “small” Delphi sample.

Efforts to establish optimum panel size have produced various results. Dalkey *et al* (1972) correlated the responses of pairs of panels of varying size and reported the resulting average statistics as reliabilities, noting an increase in reliability as group size increases from three to eleven. For 11 member panels he reported a reliability of 0.76, but omitted the number of rounds which determined this statistic. Dagenais (1978) later assembled two similar Delphi panels of 11 members each and reported reliabilities of 0.24 – 1, somewhat lower than Dalkey *et al*'s (1972), with successful convergence after round two. More recently Murphy *et al* (1998) suggest that reliability declines rapidly with groups of fewer than six participants whereas with groups of above 12 improvements in reliability are generally subject to diminishing returns. Current opinion proposes that the decision about panel size is essentially pragmatic, taking into consideration factors such as the purpose of the project, the design selected, time and expense (Hasson *et al*, 2000, Keeney *et al* 2001). Powell (2003) suggests that representation should be assessed by the quality rather than the number of panel members. Reid (1988) notes that the evidence from most studies suggests that the larger the panel size the higher the attrition rate; with panels of about 20 members tending to keep their numbers. The suggestion that having more respondents is better for reaching a consensus appears to be without empirical foundation (Murphy *et al*, 1998; Green *et al*, 1999; Hasson *et al*, 2000; Powell, 2003).

Allied to the issue of panel size is the issue of attrition over the course of the Delphi. Given the repetitive nature of the Delphi process participants can lose motivation and drop out before the last Delphi round. Critcher and Gladstone (1998) report that participation rates are typically between half and two-thirds at each stage of the Delphi. It is possible that high attrition rates can introduce a response bias which will adversely influence the Delphi findings (Williams & Webb, 1994). Walker and Selfe (1996) suggest that a minimum 70% response rate should be achieved in order to maintain rigor, although they offer no empirical justification for this particular threshold.

7.3.2.1 Size of the Expert Panel for the Study

Although there are various recommendations, there is little agreement in the literature regarding what constitutes a panel of sufficient size for a Delphi, nor is there unequivocal definition in the literature of “small” or “large” Delphi panels. In the absence of empirical evidence or criteria against which panel size can be judged other factors were considered relevant in this study. These included: ensuring the panel was of sufficient size to address the research questions; achieving maximum response and minimum attrition rates over the course of the Delphi; ensuring the Delphi could be completed in the timeframe available for the study.

The original target for the expert panel was 20 individuals, and the sampling strategy described above identified 24 experts who were eligible to participate (many names tended to recur and were identified from several separate sources). All 24 experts were approached to seek their involvement in the study, and of these 19 returned consent forms agreeing to participate [Appendix J]. Unfortunately one panelist subsequently withdrew before the study commenced citing pressure of work, and a further five, although initially returning consent forms, returned no questionnaires for any of the Delphi rounds. Thus the final panel consisted of thirteen expert members. Although comparatively small, given the homogeneous background of the Delphi subjects this still constituted a panel of sufficient size for a credible Delphi (Delbecq *et al*, 1975; Ziglio, 1996; Murphy *et al*, 1998).

7.3.2.2 Final Composition of the Expert Panel

The 13 members of the panel of experts originated from all disciplines and services providing care to children with life-limiting neurodevelopmental disabilities and their families. It was a nationally representative sample. Statutory, voluntary and charity organisations were represented.

At an organisational level six members of the panel worked in statutory services, five in voluntary services and the remaining two in independent charity services. Six members of the panel came from the general paediatric services, five from intellectual disability services and the remaining two from palliative care services. At the level of professional discipline five panelists were from nursing / therapy disciplines, three from medicine, four were service managers / coordinators, and one from the discipline of social work.

This range of expertise lends credibility to the study as it ensured a comprehensive representation of experiences, views and opinions.

7.3.3 Number of Delphi Rounds

In common with the issues of expertise and panel size the question of the number of iterations included in a Delphi also appears somewhat arbitrary. Young and Hogben (1978) suggest that the classic Delphi typically includes four rounds of questionnaires and feedback. A study by Erffmeyer *et al* (1986) provided empirical support for this, demonstrating that groups reached the point of stability in their decision making after the fourth iteration. Turoff (1975) suggests that five rounds of Delphi may be necessary to meet all of its objectives, although qualifies this by suggesting that the number may be lower where the research teams are able to formulate main issues and options with confidence beforehand. More recently Thangaratinam and Redman (2005) suggest that while decisions about the number of rounds are largely pragmatic, a valid Delphi study would consist of at least a three-iteration questionnaire. Fink *et al* (1984) propose the survey is conducted over three or four rounds, while Critcher and Gladstone (1998) suggest that the number of rounds may be as few as two or as many as five proposing that, in general, respondents' positions are unlikely to change after two or three rounds. Linstone and Turoff (1975) agree suggesting that most commonly three rounds are sufficient to attain stability in the responses with further rounds tending to show very little change and excessive repetition being unacceptable to participants. The

number of iterations in a Delphi has also been linked to the issue of attrition with Walker and Selfe (1996) and McKenna (1994a) suggesting that too many rounds may lead to fatigue by respondents and subsequently increase attrition rates. Keeney *et al* (2001) concur, suggesting that it is difficult to retain a high response rate within a Delphi that has many rounds although neither specify exactly “how many” rounds constitute “too many”.

It has also been argued that it is the achievement of response stability rather than a defined number of rounds that should herald the signal to stop polling (Scheibe *et al*, 1975; Linstone & Turoff, 1975; Rowe & Wright, 2001). Scheibe *et al* (1975) suggest this is represented by a 15% change level. They suggest that any two distributions that show marginal changes of less than 15% may be said to have reached stability, while any successive distributions with more than 15% change should be included in later rounds. Stability does not equate to complete convergence but Rowe and Wright (2001), and Rowe *et al* (2005), suggest that once response stability has been achieved it would be a mistake to conduct additional rounds in the hope of forcing consensus. They suggest that rather than a specified number of rounds designed to force consensus, response stability should herald the signal to stop polling, with disagreement accepted as informative.

7.3.3.1 Number of Rounds in the Current Study

Each member of the expert panel in the study had a busy clinical remit. McKenna (1994) suggests that this must be considered in the context of the number of Delphi iterations that it is feasible to conduct. In addition, a high number of Delphi rounds have been associated with increased attrition from studies (Walker & Selfe, 1996; Keeney *et al*, 2001) which would have been a significant issue in a study such as this with a relatively small expert panel. Consequently it was decided that this Delphi would include three iterations, and that response stability would be measured and reported as a reliability check to ensure that sufficient rounds were conducted.

7.3.4 Feedback to Panelists

Feedback between rounds is a critical feature of Delphi which enables participants to consider their position in relation to the assessments of the panel as a whole, and it has been

demonstrated that panel judgments' can be influenced by the type of feedback provided (Scheibe *et al*, 1975; Campbell *et al*, 1999). Duffield (1993) suggests that feedback can take two forms: the panel can be supplied with statistical results alone, or with a summary of comments in support of other panel members' decisions. There is some evidence that information on the reasons for divergent views is more useful than simply feeding back the ratings (Murphy *et al*, 1998; Rowe & Wright, 2001, 2005). According to Young and Hogben (1978) providing a summary of comments elicits more reasoned responses and decreases the time taken to complete a Delphi round while Duffield (1993) proposes that providing a summary of opinion also ensures that consensus is reached more quickly by two or at most three rounds. No comment is made however on the format that this should take or the increased workload for respondents.

An additional issue to be considered in the context of feedback is Jones and Hunter's (1995, 2000) warning that in feeding back the group's response it should be made clear to each participant that they need not conform to the group view, although they suggest that the researcher may ask participants who they have identified as outliers to provide written justification for their responses. Similarly Altschuld (2003) suggests that the iteration characteristics of the Delphi could potentially enable investigators to mould opinion based upon the feedback provided between rounds and advocates that the nature of feedback provided to respondents should be made explicit.

Despite its centrality to the process there is little evidence as to what is the best type of feedback, with the nature and type of feedback provided to respondents often not discussed in Delphi studies beyond a general comment that it "was provided" (Staggers *et al*, 2002; Steele *et al*, 2008; Hemmings *et al*, 2009; Green *et al*, 2009; Wilson & Moffat, 2010). Where feedback is reported it generally takes the form of measures of central tendency and dispersion although this too can take variable forms. In a study identifying the core components of cultural competence Jirwe *et al* (2009) provided respondents with the median result for each statement together with their own individual response, while in a study attempting to develop benchmarking inventories to assess the content of telephone consultations in accident and emergency departments Crouch *et al* (2002) provided respondents with the median and range for questionnaire items. In a study similar to that of Jirwe *et al* (2009), but this time identifying the core elements of nursing, Scott *et al* (2006)

provided respondents with only the mean score for each item and did not include respondents' previous scores. Mean item scores were also used to provide feedback by Snooks *et al* (2009). Conversely Haines and Critchley (2009) in a study of barriers and enablers of developing a nurse practitioner role provided respondents with bar graphs and percentages demonstrating where their opinion was situated relative to that of the group.

7.3.4.1 Feedback Strategy in Current Study

This study adopted the feedback strategy proposed by Jones and Hunter (1995) who suggest that agreement with statements be summarised and reported using the median, and consensus assessed and reported using the interquartile ranges for continuous numerical scales.

Respondents were provided with these summary statistics between rounds two and three, and because there were likely differing levels of knowledge amongst respondents with regards to the statistical analysis, a brief description of what these measures indicate was also included with the questionnaire. The mean score for rank order items was also included as this was the basis on which the ranking analysis of these items was performed. Respondents' individual scores from the previous round were also included in the feedback provided.

Comments made by respondents were not fed back between rounds as Duffield (1993) demonstrated that doing so significantly increased response times for the Delphi. However, in round three, participants identified as outliers (those in the lower and upper quartiles from the previous round) were asked to provide written justification for their views. This allowed areas of disagreement to be explored in greater depth.

7.3.5 Agreement, Consensus and Stability

The aim of consensus methods is not to force participants to come to an agreed conclusion, rather such methods provide a means of exploring whether agreement exists (Delbecq *et al*, 1975). Data from the expert panel are generally analysed using two criteria: level of agreement and level of consensus, and these can be determined both within each Delphi round and between rounds (Jones & Hunter, 2000; Greatorex & Dexter, 2000; Holey *et al*, 2007). The term agreement relates to the group's agreement with the individual Delphi

statement, whereas consensus measures the degree to which the group agrees with each other (Jones, 2002).

The criteria used to both define and determine agreement and consensus in a Delphi are subject to interpretation (Hsu & Sandford, 2007), making the issue of consensus one of the most contentious components of the Delphi method (Crisp *et al*, 1997). A universally agreed proportion for establishing agreement in a Delphi does not exist with studies recommending parameters extending from 51% to 100% (Fink *et al*, 1984; McKenna, 1994a; Sumsion, 1998; Williams & Webb, 1994; Keeney *et al*, 1996; Green *et al*, 1999; Staggers *et al*, 2002; Steele *et al*, 2008; Hemmings *et al*, 2009). Hasson *et al* (2000) suggest that the level used to define agreement and consensus depends upon several factors including the sample numbers, the aim of the research and the resources available. Keeney *et al* (2006) suggests that the answer lies with the importance of the research topic, while Fink *et al* (1984) suggest that, whatever measure is being used, the important thing is that it must be defined in advance, cautioning that the stricter the criteria, the more difficult it is to obtain agreement and consensus.

Indeed the very concept of group consensus achieved in a Delphi has been the subject of debate in the literature. It has also been argued that the move towards consensus that occurs in a Delphi reflects a normative rather than informational influence (Murphy *et al*, 1998). It is suggested that, depending upon the feedback provided, panelists may be persuaded to conform rather than express true agreement (Goodman, 1987; Murphy *et al*, 1998). In addition, Jones (2002) suggests that the mere weight of pressure to move towards the median response may cause a joint Delphi to converge towards a middle position. Witkin and Altschuld (1995) concur suggesting that subtle pressure to conform to group ratings is one of the major drawbacks of the Delphi method. Similarly Critcher and Gladstone (1998) argue that over aggressive consensus seeking may invalidate results when it is more important to explore areas of disagreement.

Scheibe *et al* (1975) suggest measuring the stability of group opinion as an alternative to consensus. They suggest that measures which take account only of the percentage of votes that fall within a prescribed range do not take full advantage of the information available in the distributions. A measure which takes account of variations from the norm is one which

measures not consensus as such, but stability of the respondents' vote distribution curve over successive rounds of the Delphi. Schiebe *et al* (1975) suggest that because the interest lies in the opinion of the group rather than that of individuals, this method is preferable to one that would measure the amount of change in each individuals vote between rounds.

However, although a critical feature of Delphi, researchers have been inconsistent in both “what” and “how” stability is measured in practice. Dajani *et al* (1979:84) define stability as “the consistency of answers between successive rounds of a study”, suggesting that stability occurs when the answers obtained in two successive rounds are demonstrated not to be statistically different from each other, irrespective of whether or not a convergence of opinion occurs. They propose the use of the Chi-square to test for group stability, suggesting that stability occurred if there was no significant difference between the response category frequencies for two consecutive Delphi rounds. However Chaffin and Talley (1980), while agreeing that the Chi-square is an appropriate test of stability, suggest that group stability is not an indication of individual response stability. Contrary to Dajani *et al* (1979), and Schiebe *et al* (1975), they advocate that it is individual stability rather than knowledge of group stability that provides more information about subsequent rounds of interrogation in a Delphi. Conversely, Holey *et al* (2007:73) cautions against the use of Chi-square as a measure of stability as it “determines the independence of the rounds from their responses and not the stability of responses between separate rounds”. Greatorix and Dexter (2000) advocate the use of graphical representations of means and standard deviations to examine stability between Delphi rounds although they specify that this is suitable only when the scale upon which experts are expressing their opinion can be considered to be an interval scale. Holey *et al* (2007) propose that stability should be demonstrated by a trend of increasing Kappa values, although again this is based upon interval level data and is applicable only when there are a number of rounds on which to measure a trend.

7.3.5.1 Definition of Agreement, Consensus and Stability in the Current Study

Both Murphy *et al* (1998) and Jones and Hunter (1995) identify two forms of “agreement” that should be considered in a Delphi: firstly the extent to which respondents agree with the issue under consideration (typically rated on a numerical or categorical rating scale), and

secondly the extent to which respondents agree with each other, the consensus element (normally assessed by statistical measures of averages and dispersion). These two criteria were applied to the analysis of data from Delphi rounds two and three in this study.

The level of agreement for each item is expressed using the median. Generally in Delphi studies, the median and the inter quartile range are more robust than the mean and standard deviation (Murphy *et al*, 1998; Hsu & Sandford, 2007), with the median less sensitive to extreme scores with small groups (Gall *et al*, 1996), and an appropriate measure to use when the data includes extreme scores, skewed distributions and / or are measured on an ordinal scale (Gravetter & Wallnau, 2008). Because the service descriptor statements presented to panelists were worded in both positive and negative terms both low and high median scored were significant in terms of the direction of panelists' agreement with each of the statements. The threshold for agreement with each statement was set at 80% as adopted by Green *et al* (1999), Staggers *et al* (2002) and Hemmings *et al* (2009).

The level of consensus in this study is expressed as the interquartile range (IQR). Consistent with other literature using Delphi and reporting these particular measures (many studies report mean and standard deviation) consensus is defined as an IQR of ≤ 1 on the five point scale used for the service descriptor statements (Anderson, 2003; Hendrix, 2005; Doughty, 2009), and ≤ 2 for the rank ordering of goals of care and priorities for change (Scheibe *et al* (1975) propose that the IQR should be no larger than two units on a ten unit scale). Disagreement, i.e. items with $> 80\%$ agreement amongst panel members, is also explored and reported.

Stability in this study is defined in accordance with Dajani *et al* (1979). Because responses are measured on ordinal scales, the sample is small, and the data are not normally distributed the use of parametric tests as advocated by Greateorex and Dexter (2000) and Holey *et al* (2007) were not considered appropriate in the context of the study. Instead non-parametric tests, as advocated by Dajani *et al* (1979) and Chaffin and Talley (1980), were used to measure response stability.

The Wilcoxon Signed Rank Test is the non-parametric alternative to the paired samples t-test. It is designed for use with repeated measures and, instead of comparing mean scores, the test

converts these scores to ranks and compares them at Time 1 and Time 2 (Howitt & Cramer, 2011). This test was used to establish group response stability between rounds two and three. The assessment of response stability was performed primarily as a reliability check as, in an effort to minimise participant burden, and due to time constraints, the number of Delphi rounds was determined a priori. The significance level was set at 0.05, and items with $p \geq 0.05$ were considered stable.

7.4 Reflection on the Application of Delphi

Delphi is a consensus method that is particularly valued for its ability to structure and organise group communication, and its capacity to bring together a wide range of expertise from different backgrounds enabling disagreement in a constructive forum which ensures equal participation by all participants. However despite widespread application in the fields of health and social care many questions regarding the application of the Delphi method continue to intrigue researchers. In general, criticism of Delphi arises from the fact that there are no standards established in any methodologically acceptable way, with the method criticized on the grounds of reliability measurements and a lack of scientific validation of the findings (Sackman, 1975; Reid, 1988; Willaims & Webb, 1994). However it has also been counter-argued that Delphi's significant use in resolving situations where no definitive evidence is available, and its application in areas that do not lend themselves to traditional scientific approaches, may mean that it is not appropriate to use the same validation criteria as for hard science (Fink *et al*, 1984). Critcher and Gladstone (1998) propose that at a philosophical level Delphi is a victim of its hybrid epistemological status. They suggest that while Delphi may provide quantified results within a recognisable positivist tradition, the definition of the problems and their solutions by those who are the subjects of the research place it close to constructivist positions. Mullen (2003) concurs, suggesting that many of the relatively recent criticisms of Delphi stem from the positivist approach whereas Delphi studies straddle the divide between qualitative and quantitative methodologies (Stewart, 2001).

Although the method has been criticized for its many hybrid forms and variations in format it is perhaps this adaptable and flexible nature which has contributed to its widespread implementation by practitioners and researchers. Despite the methodological criticism,

Delphi's effectiveness over comparative methods, at least in terms of judgmental accuracy, has generally been demonstrated, with research suggesting that Delphi allows improved judgment compared to alternative methods (Rowe & Wright, 1999; 2001). In addition Delphi's aggregation method is explicit thus enhancing the rigor of the technique (Murphy *et al*, 1998).

Although not without its critics, Delphi appeared to be the most appropriate method for this phase of the study based upon the purpose of the phase and the research questions to be addressed. The rationale for choosing Delphi has already been outlined in the introduction, and the method has been widely used in similar aspects of healthcare research for the identification of clinical and research priorities and the validation of practice (Scott *et al*, 2006; Carnes *et al*, 2010; Chang *et al* 2010; Sue Hoyt *et al*, 2010; Jeste *et al*, 2010; MacNeela *et al*, 2010, West, 2010; Wilson *et al*, 2010; Wilson & Moffett, 2010; Zeigler & Decker-Walters, 2010). The value of Delphi in the context of this study was its ability to generate ideas and insights, those that evoked agreement and consensus and those that did not, from expert clinicians for whom it would have been impossible to organise mutually agreeable times and places for meetings. Additionally the use of a qualitative technique to draw on collective expert judgment in a format that allowed for subsequent quantitative analysis of these data was compatible with the overall mixed methods approach used in the study.

The previous section has discussed the critical features of a Delphi study and their application to the current study. The remainder of this chapter discusses the conduct of the study and presents the findings.

7.5 Data Collection and Materials

This Delphi proceeded in two stages: the first stage involved the conduction of three preliminary interviews with expert practitioners which are described below. The second stage involved the use of self-report postal questionnaires to the thirteen members of the expert panel. In each round participants were issued with the relevant questionnaire and a letter outlining the process to be followed in its completion. Since this process necessitated the use of a variety of data collection instruments, the following section describes the development of these instruments in the chronological order in which were used in the study.

7.5.1 Preliminary Interviews

In order to accumulate as much cogent and relevant information as possible in advance of formulating the questionnaires to be administered in the iterative Delphi rounds, three semi-structured interviews were conducted with experts whose selection has previously been described. The interview topic guide centered on the research questions with the subject related topic areas identified a priori [Appendix T]. Participants were asked to discuss what they considered to be the goals of care for this population of children and their families, how well they felt current services meet the needs of these children and their families, and what changes they felt were required to improve services to the population. These guiding questions were supplemented and supported by prompts and probes when appropriate to encourage participants to expand upon an answer, or to redirect them back to the main topic if the focus became lost. Interviews were conducted at a time and place convenient for participants, and were tape recorded.

7.5.2 Round One Questionnaire

Delbecq *et al* (1975) propose that developing the question / problem statements is the key to the Delphi process. They suggest that if respondents do not understand the initial broad question they may answer inappropriately or become frustrated and lose interest in the questionnaire with potentially negative effects on the quality and quantity of responses. Issac and Michael (1995) describe the function of the first Delphi questionnaire as the generation of items, issues or questions.

In the first round of this Delphi participants received a cover letter [Appendix L] and a questionnaire containing only open ended questions [Appendix M]. The use of an open-ended questionnaire in round one ensured that all participants were allowed the freedom to bring their own views to the study. It allowed panelists to specify the key issues to be addressed rather than compelling them to answer researcher-developed questions that they may have felt were unbalanced, incomplete or irrelevant. Lemmer (1998) suggests that this is an important aspect in the context of developing a comprehensive questionnaire as it ensures representation of all views at the outset. Allowing individuals input from the outset can also help prevent attrition in subsequent rounds (Keeney *et al*, 2006). Payne (2004) identified several advantages to the use of open-ended questions including that they are uninfluenced;

elicit a wide variety of responses; provide background for interpreting the answers to other question; can be used to solicit suggestions, obtain elaboration, elicit reasons and evaluate arguments; and are of value as a preliminary aid in drafting other questions especially in preparing questions on an unexplored issue.

The questionnaire was structured around the study's research questions. It clearly defined the terms used in the study and elicited panelists views on the broad subject areas related to the focus of the study. Panelists were asked to indicate (1) what they considered to be the goals of care for children with life-limiting neurodevelopmental disabilities and their families, and to explain how / why they believed each goal to be important; (2) to describe which goals they felt are achieved in the context of current services and to indicate what they believe is preventing the achievement of others; and (3) to identify changes to current services they considered would improve the care provided to the population and the resources that would be required in order to achieve these changes. Questions 1 and 3 related specifically to the first and third research questions. Data from the second question was used to assist with the development of the problem statements / service descriptors used to address the second research question.

The questionnaire was reviewed by the three expert interviewees for face and content validity before being distributed to the expert panel.

7.5.3 Delphi Questionnaire for Rounds Two and Three

Previous Delphi studies cite several potential sources for the identification of items for the Delphi questionnaire. While Staggers *et al* (2002) developed their statements directly for the literature, more commonly researchers have used a qualitative approach, including individual and focus group interviews, and open-ended questionnaires to develop the items for the iterative rounds (Scott *et al*, 2006; Jirwe, 2009; Green *et al*, 2009; Wilson & Moffat, 2010). This study used the data returned in the questionnaire from Round One, combined with the expert interview data. This combined data set was used as the basis for developing the problem statements / service descriptors used in the questionnaire for Rounds Two and Three.

The second and third round questionnaires were structured in nature, presenting respondents with a list of statements and closed ended questions [Appendix U]. The instrument was structured in three sections related to the three research questions to be addressed. In the first section participants were asked to rank order the goals of care that had been identified in round one (1 = most important), and were given the opportunity to include textual comments in support of their position if they desired.

In the second section participants were asked to score their agreement with service descriptor statements using a five point Likert scale with responses ranging from 1 – *Strongly Disagree* to 5 – *Strongly Agree*. The statements in section two of the questionnaire were categorised into six distinct areas which had been identified in the interview data and the responses from Round One. These included statements related to the structure and funding of services; services available to children and their families and the process of their delivery; the integration and coordination of services; issues related to the provision of palliative care; ease of access to services; and working in partnership with parents. Respondents were again given the opportunity to comment on individual statements and invited to include any additional comments they wanted in free text at the end of each section.

Section three asked participants to rank, in order of their potential to improve care, the service-related changes that they considered were most important to improve services to children with neurodevelopmental disabilities and their families (1 = most important change).

7.5.3.1 Piloting the Delphi Questionnaire

Okoli and Pawlowski (2004) suggest that pretesting, or piloting, of the instrument is an important reliability assurance for the Delphi method. Because of the small panel size expert members of the Delphi panel were not used to pilot the data collection instruments for this study as this would have further reduced the size of the expert group. Instead the expert interviewees were asked to pilot the questionnaires. No changes were made to the instrument following the pilot stage. The pilot questionnaires were not used in the data analysis.

7.6 Data Analysis

The following section discusses the analysis of data obtained in the Delphi. Because of the multiple stages in the procedure, and the collection of both qualitative and quantitative data, the analysis of data is described separately and sequentially below.

7.6.1 Analysis of Interview Data

The three preliminary interviews were transcribed verbatim into a Microsoft © Word document and analysed using a simple content analysis. Julien (2008:120) describes qualitative content analysis as “an analytic method of reducing and making sense of data”. The purpose of data analysis in this case was to ensure a comprehensive and representative range of views that could be included in the development of the questionnaire for the iterative rounds of the Delphi. In this respect the interview data were not expected to stand alone as an in-depth analysis and discussion of the issues, but rather to augment the findings from the open-ended questionnaire used in round one and ensure the inclusion of all relevant issues. Consequently a surface rather than deep-level analysis of interview transcripts was conducted.

The first stage of analysis involved familiarization with the data corpus. All interviews transcripts were repeatedly read so that the overall breath, depth and content were familiar. The second stage involved the analysis of individual interview transcripts. Each transcript was analysed line by line with the extraction of text related to each of the three topic areas (Goals of Care, Service Related Issues, and Priorities for Service Improvements). Extracted text was then pasted into separate documents and each document was examined individually. An example of this analysis is presented in Appendix V.

The “Goals of Care” and “Priorities for Service Change” described by interviewees were listed and counted. The majority of interview data related to interviewees’ perceptions and experiences of the general structure and functioning of services. Several separate aspects of services were evident in the data when coded, these included issues related to the structure and funding of current services; the process of delivery and the services available to families; integration of services; ease of access to services; orientation of services towards parents; and issues related to palliative care for the population.

Text related to each of these aspects of services were extracted and condensed into service descriptors (i.e. a statement related to some aspect of service provision). These service descriptors were presented as statements to panelists in Round Two with panelists asked to indicate their level of agreement with each service descriptor statement on a five-point Likert scale.

The process of identification of goals of care, service descriptors, and priorities for service changes is discussed further in the findings section of this chapter.

7.6.2 Analysis of Round One Questionnaire

Payne (2004) suggests that the major difficulty with open-ended survey questions relates to the coding and quantification of answers, while Campbell (2004) cautions that the freedom of response that open-ended questions permits respondents can lead to ambiguity rather than clarity. Hasson *et al* (2000) propose that data returned in the first round questionnaire can be analysed using simple content analysis. Subsequently the simple content analysis procedure used to analyse interview transcripts was repeated and used to summarise the panels' responses to the open-ended questions from Round One. The purpose of this round was to generate as many perspectives on the focus areas as possible, and to categorise responses in a manner that could be quantitatively analysed.

For Question 1, which related to the identification of goals of care for children with life-limiting neurodevelopmental disabilities and their families, this involved arranging all responses in a single list. The list was carefully reviewed for overlap and duplicate content, and then combined with the interview data related to the same area. Following the elimination of redundant items (i.e. exact duplicates) the identified goals were subsequently listed in the second round questionnaire where respondents were asked to rank the goals in order of their importance. Where possible the exact wording provided by the panelists in Round One was used, but in the case of ambiguous or multiple responses related to the same goal the clearest and most succinct statement was selected for inclusion in the second round questionnaire.

Question 2 asked respondents to identify which, and why, goals identified were not achieved in the context of current services. This data, in tandem with the corresponding interview data, formed the basis of the service descriptor statements for the second and third round questionnaires. Again the text was carefully reviewed for overlap and duplicate content removed. This textual data was then combined with the interview data and the procedure repeated. Descriptor statements were extracted from the combined data sets, and were developed to represent the concepts contained in the data rather than an exact replication of the panelists wording. Examples of the procedure used to develop service descriptor statements are presented in Figure 7.3.

Figure 7.3 *Combining Data Sets to Develop Statements for Iterative Rounds*

Interview Data	Response from Round 1
And I think like we know the parents deliver the care... but I don't think we include them sometimes... in discussion around care....and I do think that would be a great benefit for families.	Doctors are very slow to take on board parents' experiences of how the child is. I feel parents are not listened to enough..... and we all know parents know their child best..... They live with their symptoms 24 hours a day....
Service Descriptor Statement Extracted	
<i>Parents are considered equal partners in the setting and prioritising of goals for their child's care.</i>	
Interview Data	Response from Round 1
I mean I do know here at times I would look for medical teams to meet together with the family...em so that.... so that they know eh, that they do have a sense that everybody is looking at the greater child's needs rather than em the individual or the individual sort of symptom or problembut it is difficult....	Parents often have to deal with numerous agencies, i.e. the hospital, acute setting, disability services, respite provider plus HSE disability service. These children are very complex and have many needs. Would benefit from improved multi-agency working to address needs.
Service Descriptor Statement Extracted	
<i>It is difficult to get a holistic view of the child's needs because of the number of different health professionals and services involved in the child's care.</i>	
Interview Data	Response from Round 1
a key worker can be a huge benefit... you know somebody that can grab all of the different potential teams out there.... or services out there.... get them together and say "right look what can we all do, lets co-ordinate a care plan around this child.... it can work very well....	Caring for a child with a life-limiting condition can be hugely stressful for parents, a family-support worker / nurse becoming involved in the child's home is essential to assist the family.
Service Descriptor Statement Extracted	
<i>The lack of a key worker for families results in the ad hoc delivery of services to this population.</i>	

Analysis of Question 3, the proposed changes to services that would improve care for this population, was analysed in the same way as Question 1. The service-related changes that were identified were listed in the second round questionnaire where respondents were asked to rank order the proposed changes in terms of their potential to improve care for the population. Again, where possible, the exact wording provided by the panelists in Round One was used.

7.6.3 Analysis of Round Two and Three Questionnaires

Quantitative data from Delphi rounds two and three were analysed using SPSS® version 17. In this study measures of agreement and disagreement are represented by the median score, with a threshold set at 80% for service descriptor items. Consensus is represented by the interquartile range [IQR]. The justification for using these particular measures has already been provided. In addition, mean scores were also calculated for “goals of care” and “priorities for service-related changes” in order that these items could be ranked in order of the priority awarded to them by the panel as advocated by Wiener *et al* (2009).

7.6.3.1 Consistency of Questionnaire between Rounds Two and Three

Opinion on the consistency of Delphi questionnaire items from round to round is variable. Critcher and Gladstone (1998), Green *et al* (1999), and Wilson and Moffat (2010) reported eliminating infrequently occurring or low scoring items from round one of the study in order to keep the resulting list more manageable. Endacott *et al* (1999), Staggars *et al* (2002), Steele *et al* (2008) and Jirwe *et al* (2009) removed questions on which consensus had been reached in Round Two from subsequent Delphi rounds. However Hasson *et al* (2000) and Hsu and Sandford (2007) caution against such decisions suggesting that all items are included in each round as their removal goes against the basic tenets of the Delphi technique. Similarly Hemmings *et al* (2009) included all items in each round of a three round Delphi.

This study included all items in all Delphi rounds, including those on which agreement and consensus had been reached in Round Two. The reason for this was twofold: firstly it ensured that comprehensive feedback on all items was provided to the expert panel; secondly, because the number of iterations for the study was decided in advance, including all items in

Round Three was necessary in order to assess response stability. This was important in the context of ensuring that a fourth Delphi round would not have resulted in significantly different findings.

7.6.3.2 Management of Non-Responses and Missing Data

The literature proposes several strategies designed to maximise response rates and minimise attrition all of which were employed in this study. These included deciding in advance the number of Delphi round to be conducted in order that panelists would know exactly what time commitment was involved (Walker & Selfe, 1996; Citcher & Gladstone, 1998; Keeney *et al*, 2001); where possible personally approaching individuals to invite participation before commencing the study (McKenna, 1994a; 1994b; Vernon, 2009); and providing the panel with sufficient information before commencing the study (McKenna, 1994a; Hasson *et al*, 2000). The layout of the questionnaire and clarity of questions were piloted in advance of distribution to the panel members (Hatton & Nunnelee, 1995). All questionnaires included a defined date for completion, with a reminder letter posted to each panelist who did not return a completed questionnaire by the specified date for each round (Salmond, 1994).

The management of non-responders is also variable in the literature. While some authors provide no discussion of this issue (Scott *et al*, 2006; Wilson & Moffat, 2010), others have handled it in opposing manners. Butterworth and Bishop (1995), Staggers *et al* (2002) and Steele *et al* (2008) included only those who returned completed questionnaires in subsequent Delphi rounds. Conversely Endacott *et al* (1999) and Wiener *et al* (2009) included non-responders from previous rounds.

This study adopted the strategy described by Endacott *et al* (1999) and Wiener *et al* (2009). All members of the expert panel were included in all Delphi rounds regardless of whether they had returned a completed questionnaire in the previous one. This decision was a pragmatic rather than philosophical one. The expert panel was small to begin with; excluding non-responders from subsequent rounds would have resulted in a significant reduction in panel numbers which would have severely limited the validity of the study. In addition, on both occasions where the cause of non-response has been investigated in previous studies, this was found to be associated with pressures of work (Butterworth & Bishop, 1995;

Zollingen & Klaassen, 2003). It was considered punitive to exclude busy clinicians from the panel on the basis that they did not have the time to complete one round of a three round questionnaire. Subsequently all missing data (non-returned questionnaires and returned questionnaires with missing values) were entered into SPSS as missing data.

7.7 Findings

The following section presents the findings of this Delphi. The findings from the preliminary rounds (Interviews and Round One) will be discussed separately as these were distinct and separate data collection instruments. However, as their purpose was not to stand alone as independent findings these preliminary findings will be discussed only briefly. The primary focus of the section will be on the findings from Round Three of the Delphi as this represents the final opinion of the expert panel, and the changes that occurred between the second and third rounds. Consequently rounds two and three will be discussed together so that comparisons can be made. The section begins with a discussion of the interview findings as this was a separate group to the main expert panel.

7.7.1 Interview Findings

The goals of care for children and their families identified from the interview data related to three distinct areas: symptom control and quality of life, the provision of appropriate support to parents, and the provision of a seamless web of care to the child and family. Both symptom control and quality of life, and the provision of appropriate support to parents were all cited as important goals of care by all interviewees, while the provision of a seamless web of care arose in one interview. Examples from the interview transcripts and corresponding goals of care are presented in Figure 7.4.

Figure 7.4 *Interview Excerpts and Corresponding Goals of Care*

Interview Excerpt	Goal of Care
I suppose I am naturally biased towards symptom management and symptom control..... and I think for a lot of these children that's really important [Int 01] "I suppose they really from my opinion they would be optimising the quality of life of that child..... I suppose you could look at in terms of their physical needs, their spiritual needs of the child and family, the psychosocial needs. They are the primary goals in my opinion" [Int 03].	Optimising quality of life for the child
"your goal is providing maximum comfort for the child... and support is one of the biggest goals I would think... for the child and the family"...[Int 02] "you have to respond to families needs so.....em... you know if you have a parent in distress you have to respond to that distress" [Int 01]	Providing support for parents
"and its hospital care and community care and home care you know... and you're trying to coordinate all that seamlessly.....you know so that it works smoothly....is nearly is what you are trying to do..."[Int 03]	Achieving a seamless web of care

The goals of symptom control and quality of life were linked in the interview data, for example

"I think that one of the biggest things in setting goals of care is that we always provide the family with a hope that things may get better....that we can get on top of symptoms, or that the child is going to live a good life...." [Int. 02]

Providing "support" to the family was cited by all interviewees as an important goal of care. Although this support took different forms (informational support, psychosocial support, providing hope and encouragement) it served a consistent function which was to make life "easier" or "simpler" for the family. Achieving a seamless web of care was considered important by one interviewee. This concept related to a smooth integration of all the services involved in the care of the child whether these were hospital, community, or home based. All three of these goals were reiterated in the open-ended questions of the first Delphi round, and were carried forward and listed to be ranked by panelists in Round Two.

The majority of interview data related to interviewees' experiences of the general structure and functioning of services. Six aspects of current services were identified upon repeated re-reading of the interview transcripts. These included: the structure and funding of services; the services available to the population and the manner in which they are delivered; the integration of services; access to services; services' relationship with parents; and issues associated with the provision of palliative care.

In some areas specific issues associated with acute and community services were separately identified, while in others there was a global focus in relation to coordination, integration and collaboration of services generally. Issues related to the structure and funding of services were identified in two interviews. This related to the geographical inequality of community based services whereby the availability or lack of a service depended not upon the level of need of the child, but upon where the family was resident. Inconsistent financial resources were also identified as a cause of the unequal distribution and allocation of services, even within the same geographical area.

In relation to the services available to the population all interviewees identified inadequate provision of psychological support to parents as a major problem. Interviewees proposed that families experienced considerable levels of psychosocial need, in some instances equaling the physical needs of the child. Psychological stress was identified as an ongoing problem for parents, but one which could be easily overlooked in current services

“There needs to be acknowledgement by medical people that parents are under as much psychological trauma as they are physical with their child’s care....and I suppose personally I would feel at times meeting the psychosocial needs of families is extremely difficult and not always possible”.... [Int. 01]

The orientation of the services also arose as important features in the interviews. All interviewees stressed that care of a young child with a life-limiting neurodevelopmental disability should be orientated towards the family as a unit, i.e. should be family focused. It was considered critical that the child should be seen in the context of the family, which one respondent described as “child orientated, but family focused”. [Int. 01]

One community-based interviewee identified a conflict between the medical model of care (relevant to the child’s physical needs), and the social model of care (prevailing model in disability services), expressing the opinion that, especially as children get older, this can cause a significant problem in terms of attending to the complex medical needs of the child.

Integration and coordination of services arose as an important issue for all interviewees. All expressed the opinion that services are poorly integrated, both at the level of the different health-professionals involved in the care of the child, and more generally at the level of the

multiple services involved in the care of the child. Interviewees attributed this to poor communication between health-professionals, and to poor communication between services, and the lack of available key-workers

“certainly from my experience in the hospitals I have worked in...we’re not very good as professionals working sort of interdisciplinary wise...or having effective communication...its very much nurses doing their nursing bit, the doctors doing the doctor bit, the physios do their bit, or one specialty looks after that part of the child, another specialty concerns themselves with neurological issues...and were not effectively communicating..... and if we can’t effectively communicate amongst ourselves, it is very difficult for us to be able to effectively communicate with a child and family” [Int. 02]

Services relationship with parents also arose as important features in the interviews. All interviewees proposed that parents were the experts in the care of their particular child. Parental expertise was considered important to the holistic care of the child in that it provided a picture of the whole child rather than the specific individual problems experienced. Despite this it was considered that services often exclude parents from decision making and goal setting for the child.

Issues associated with the delivery of palliative care arose in two interviews. Both of these interviewees were involved in the care of young children with life-limiting neurodevelopmental disabilities in the context of hospital based services. It did not arise as a feature in the third interview conducted with a community based health-professional. Both interviewees suggested that a palliative care approach to children with life-limiting neurodevelopmental disabilities and their families is important, although it was emphasised that specialist palliative care is not required by all of the children in this population. Interviewees stressed that the important feature of specialist palliative care was that it should be available and easily accessible if, and when, it is required.

Statements related to each of these areas were extracted from the transcripts and resulted in the development of 22 service descriptor statements for Round Two.

Seven changes to services were identified that would improve care to these children and their families. The following categories were apparent in the data: changes required to improve

acute and community services; improved overall coordination of primary and tertiary services; the need for a parent held record; and the need for a key worker for every family. The proposed changes identified by interviewees and supporting interview excerpts from interview transcripts are presented in Table 7.5.

Figure 7.5 *Interview Excerpts and Corresponding Proposed Changes to Improve Services*

Interview Excerpt	Proposed Improvement to Service
“if you list the number of people involved with our service in all there could be 7 or 8 different professionals, then if that child is attached to the hospital, then there is the various consultants, and bearing in mind they could be attending 4 different consultant appointments that are not necessarily in communication with each other”.... [Int. 01]	A greater level of communication between the different health professionals involved in the care of the child.
“and one in particular that I have been in contact with recently had nearly co-opted me as a key worker....in terms of networking...in terms of “this is what I need at the moment”... they will say to you “I cannot face making this call, because there are four or five things that I need, and I’m not going to be able to cope with four or five different things at the moment”...so I really feel they lack a gate you know...it’s very overwhelming for them... plus the amount of professionals coming and going”....[Int. 02]	A key worker available to every family.
“I think we should have better hand held notes....I mean most parents have notes that they have made up themselves...which are fantastic, but I think if we were to medicalised them a bit more....so that you know regardless of what service your are, you know that this family is going to have a passport, or whatever you want to call it...and you know that on the 3rd. page is going to be their medication you know....and on the 4th. part of the folder is going to be their last medical letter”....[Int. 01]	Parent held medical records.
“because quite often the acute setting doesn’t know what the primary care setting is doing...so there needs to be a bit more of a structured approach....more coordinating the services”... [Int. 03]	Improved liaison between acute services and other services involved in the care of the child.
“So it would be access to service when they need it... but they may not need special palliative care all of the time, but when they do need it and for the period of time that they need it that they can have access to that service where it is suitable to them”... [Int. 01]	Access to specialist palliative care in a timely and efficient manner
“if you could grab all of the different potential teams and services out there... get them together and say “right, look what can we all do...let’s coordinate one care plan around the child....that could work very well”... [Int. 03]	A single care plan that is used across all services.
“there are still families being discharged home without any services set up for them...where in fact there are very good services in the community....but maybe because the nurse at the time wasn’t aware of something....so I think a review of the services in the community so that there is nearly a directory of knowing who is where, what they provide and what their admission criteria are...[Int. 03]	A national directory of services available to children and their families.

Acute services changes were primarily focused on aspects of greater coordination of clinics and appointments to reduce the burden of travel on families, and awarding “priority” status to

these children in clinics and accident emergency departments. Community service changes proposed related to the establishment of small specialist community based teams¹¹ who would attend to the needs of the child and family in the community setting. All interviewees identified the requirement to improve the overall coordination of primary and tertiary services. It was suggested that the establishment of a central coordinator would ensure that services were appropriate to the changing needs of each individual family, and also ensure that resources were equitably and fairly distributed amongst the population who need them.

The need for a parent-held record was also identified by all interviewees. It was proposed that this had the potential to improve care in a variety of ways. These included improving communication between health professionals; improving communication between services; providing valuable knowledge about the child as a person; and alleviating the burden of repetition of information on parents. All interviewees additionally emphasised the requirement for a designated key-worker for every family, and considered a key-workers potential for improving care to families was immense. This included reducing the burden for parents by making it easier to access what the child needs, facilitating access to acute services when appropriate, coordinating all of the services and agencies involved in the child's care, alleviating parents worries and fears and providing a sense of support for parents. All seven proposed changes to improve services were carried forward to be ranked by respondents in Round two of the study.

The remainder of this section will focus on the iterative rounds of the Delphi and the opinion of panel of experts.

7.7.2 Expert Panel Response Rates

Response rates for individual Delphi rounds were variable ranging from 69% to 100% [Table 7.1] with 46% of panelists (n = 6) completed all three rounds.

¹¹ This was specified as a “core” specialist team. The establishment of specialist paediatric palliative care teams was not identified as a priority in the interviews.

Table 7.1 *Response Rates for Individual Delphi Rounds*

Delphi Round	No. of Respondents	% Response Rate
Round 1	9	69%
Round 2	13	100%
Round 3	12	92%

n for each round = 13

Although the response rate for round one was 69%, examination of non-responders indicated that they were spread amongst all the subgroups. This eliminated the potential for bias that may have occurred with more systematic non-response. This, in association with the fact that there was a large amount of data gathered in the qualitative interviews, ensured that there was sufficient expertise amongst the sample to allow the study to proceed with the development of the structured questionnaire for rounds two and three.

7.7.3 Findings from Delphi Round One

Nine individuals (69%) returned completed questionnaires for round one. This was the lowest response rate for any of the rounds in this Delphi, possibly as, according to Bourque and Fielder (1995), respondents are generally reluctant to answer open-ended questions in a self-administered questionnaire.

The expert panel returned 13 goals of care for young children with life-limiting neurodevelopmental disabilities and their families in this round. Three of these had already been identified by interviewees. Although the issues of symptom control and quality of life had been linked by the interviewees this was not the case in the open-ended questions where these were listed as separate goals by most respondents. Subsequently they were returned separately to respondents to rank order in Round Two. The goals of care identified in this round, and the frequency of their occurrence are presented in Table 7.2.

Table 7.2 *Goals of Care Identified in Round One and Frequency of Occurrence*

Goal	No. of Times Identified
The child is cared for at home	4
Achievement of the best possible quality of life for the child	6
Provision of appropriate respite	4
Achievement of the child's full potential within the limits of the illness	4
Inappropriate medical interventions are minimised	2
The family continues to function as a unit and enjoy life	1
Promotion of normality as much as possible for the child and family	3
Open and honest communication with the family	1
The child's life is prolonged	1
Optimum management of symptoms	5
Parents are supported with the provision of care	9
The family is provided with the hope that things will get better	1
Achievement of a seamless web of care	4

n = 9

The issues identified by interviewees in relation to service provision to this population of children and their families were reiterated by the expert panel in the first Delphi round. Therefore the original coding frame and twenty-two service descriptor statements developed from the interview data remained valid. However other issues were identified by panelists that had not been identified in the interviews. These were related to the issues of in-home support and respite; a more expansive concept of palliative care; a lack of expertise amongst some health-professionals; and a heavy reliance upon charity services to meet the needs of the population.

Some panelists indicated that they considered home to be the ideal place of care

Home is the best place to care for all children, regardless of disability. All children have a right to be cared for at home [05]

In-home support was considered was considered vital to continued caring as

Parental exhaustion and burnout is common where home support is inadequate [07]

Despite this many reported that there was insufficient in-home support for families caring for young children with life-limited neurodevelopmental disability, particularly in relation to end-of-life care.

Some panellists specified additional resources that were required for in-home support, for example home-help, additional nursing hours, and access to essential equipment. Others emphasised the critical role played by respite when facilitating families to continue to care for their child at home. Respite could be provided in or out of the home, but panellists' perception was that current respite facilities are insufficient to meet the needs of parents.

Although issues of access and availability of palliative care services arose in the interviews a more expansive concept of palliative care arose in panelists' open-ended responses in Round One. This related more to the overall approach to care for young children with life-limiting neurodevelopmental disabilities and their families rather than access to a specific service. Panellists proposed that care for these children "should not be over medicalised" [07], and opined that currently "many children undergo futile investigations and procedures" [04]. There was the perception that sometimes over aggressive management of the child's condition could result in unnecessary suffering. Panellists associated this with a lack of meaningful communication between the health-professionals involved in the child's care and the child's parents

Sometimes there is a failure to discuss "life-limited" honestly with parents – so medical staff continue to adopt a very active management approach to the detriment of the child [05]

Panellists also reported a perceived lack of education on the part of some health-professionals. While it was considered that all health professionals caring for young children with life-limiting neurodevelopmental disabilities and their families should have specific education and training to meet their needs, General Practitioners were specified as a particular group in need of additional education particularly in the context that "G.Ps are not paediatricians" [05]

The final issue to arise in open-ended responses that did not feature in the interview transcripts was the perception that currently there is a heavy reliance upon charity services to meet the needs of the population. Analysis of the textual responses from Round One resulted in the addition of 12 new items to the list of service descriptors. This brought the final number of statements on which panelists were asked to rank their agreement in Round Two to thirty-four.

Eighteen separate service-related changes were identified from textual responses in Round One (including the eight already identified by interviewees). All eighteen were carried forward to Round Two to be ranked in order of their potential to improve services to this population of children and their families. The proposed service related changes, and the frequency of their occurrence, are presented in Table 7.3.

Table 7.3 *Proposed Changes to Services and the Frequency of their Occurrence*

Change Required to Current Services	Times Identified
A greater level of communication between the different health professionals involved in the care of the child	1
A key worker available to every family	3
A single care plan that is used across all services	1
A greater level of coordination and integration of all services involved in the care of the child	1
A single point of contact for information for families	1
Less beurecracy surrounding the family's entitlements	1
Access to specialist palliative care in a timely and efficient manner	1
Parent held medical records	1
A national directory of the services available to children and their families	1
Improved education of community based health professionals	4
A specialist paediatric palliative care consultant to act as a resource when required	2
A formal coordinator of services for children with life-limiting disabilities in every HSE area	1
Affording children "medical priority" status in Emergency and Outpatient departments	2
Development of community based paediatric palliative care teams	4
Improved respite facilities	3
Less protracted ordering system for essential equipment	2
Improved liaison between hospital services and other service providers	3
National standards of care and services	1

n = 9

7.7.4 Findings from Delphi Rounds Two and Three

The previous sections described the findings from the qualitative analysis of the expert interviews, and the findings from Rounds One of the Delphi questionnaire. However the principal focus in a Delphi study is on the agreement, or lack of agreement, achieved in the final Delphi round and the stability of response between rounds.

This section presents the findings from Delphi Rounds Two and Three. The principal focus will be on the findings from the final round, and the changes that occurred between the rounds. Each of the topic areas that the Delphi aimed to investigate will be presented separately. The section begins with the findings related to establishing the goals of care for young children with neurodevelopmental disabilities and their families. This is followed by experts' opinions on the current services available to this population of children and their families and the process of service delivery. Finally expert opinion on the proposed changes that would improve services to this population of children and their families are presented. In order to facilitate a comparison of the changes that occurred between rounds the results of rank ordered items (goals of care and proposed changes to services) are each presented in a single table for rounds Two and Three.

Thirteen completed questionnaires (100%) were returned in the second Delphi round and 12 in the final round (92%).

7.7.4.1 Goals of Care

In Round Two the panel agreed that achieving of the best possible quality of life for the child was the most important goal of care as indicated by the lowest mean score and achievement of consensus amongst the panel (IQR = 1) [Table 7.4]. “The child is cared for at home” was awarded second priority, although the IQR = 4 indicated less agreement amongst the expert panel with regards to the priority awarded to this goal. The lowest priority was awarded to “the child’s life is prolonged” and “the family is provided with the hope that things will get better”. This position is consistent with a palliative approach to the care of the child and family. Both of these goals also achieved consensus ($IQR \leq 2$) indicating agreement amongst the panel that these were the least important goals with regards to this population of children and their families.

There was relatively little change to the rank order assigned to the priority awarded to goals of care between rounds two and three [Table 7.9]. Five goals retained their priority status from round to round, while an additional seven goals moved up or down one ranking place in Round Three. The priority status of the goal “The child is cared for at home” demonstrated the greatest shift between rounds falling from second place in Round Two to fifth place in Round Three. However, Wilcoxon Signed Rank Test demonstrated that, despite the change in priority status, there was no significant difference in the mean scores for this goal between rounds Two and Three ($W = .00$, $p = 1.00$).

Table 7.4 Respondents Rank Order of Goals of Care in Rounds 2 & 3

Goal	Round 2 Priority	R2 Mean	R2 Median	R2 IQR	n	Round 3 Priority	R3 Mean	R3 Median	R3 IQR	n	W	p
The child is cared for at home	2	4.15	4.00	4.00	13	5	4.08	3.00	2.50	12	.00	1.00
Achievement of the best possible quality of life for the child	1	1.92	1.00	1.00	13	1	1.17	1.00	0.00	12	-1.34	.18
Provision of appropriate respite	8	6.62	7.00	5.00	13	8	7.25	7.50	3.50	12	-.45	.66
Achievement of the child's full potential within the limits of the illness	4	5.15	5.00	6.00	13	3	3.90	3.50	3.00	12	-1.60	.11
Inappropriate medical interventions are minimised	11	8.00	8.00	4.50	13	10	8.58	9.50	3.00	12	-1.22	.22
The family continues to function as a unit and enjoy life	9	6.85	5.00	5.00	13	9	7.75	8.50	3.75	12	-2.03	.04
Promotion of normality for the child and family	6	6.08	5.00	6.00	13	7	6.83	6.00	3.75	12	-1.07	.29
Open & honest communication with the family	3	4.38	4.00	2.50	13	2	3.75	3.00	1.75	12	-1.60	.11
The child's life is prolonged	12	11.54	12.00	1.00	13	12	11.50	12.00	0.75	12	.00	1.00
Optimum symptom management	5	5.69	5.00	5.50	13	4	4.00	4.00	2.00	12	-1.86	.06
Parents are supported with the provision of care	7	6.54	6.00	2.50	13	6	6.75	7.00	2.75	12	-1.34	.18
The family is provided with the hope that things will get better	13	11.69	13.00	1.00	13	13	11.83	13.00	1.00	12	-1.34	.18
Achievement of a seamless web of care	10	7.54	7.00	2.50	13	11	8.67	9.00	3.75	12	-1.60	.11

1 = Most Important Goal
Shaded cells indicate consensus

Despite there being some changes in order, the five highest ranking goals from Round Two retained their top five ranking in Round Three, with three of these achieved consensus in round three. These included: “achievement of the best possible quality of life for the child”, which retained it’s number one priority between rounds; “open and honest communication with the family”, which increased from third priority in Round Two to second priority in Round Three; and “optimum symptom management”, which increased from fifth priority in Round Two to fourth priority in Round Three.

Two of the top five goals of care identified by the expert panel failed to reach consensus in the final round. Although ranked as the third highest priority goal “achievement of the child’s full potential within the limits of the illness” did not achieve consensus amongst the panel (IQR = 3). Three panelists provided textual commentary in respect of this goal. For two panelists the goal was synonymous with access to appropriate education and was inextricably linked with the issue of overall quality-of-life for the child

“Again this is a quality of life issue. Community services can be under resourced and unable to provide pre-schooling / schooling to children with more complex needs” [ID. 02], the third panellist offered a different perspective, proposing instead that a focus on this goal accrued benefits for the family rather than the individual child, suggesting that

“it is very important to always give the family hope and an achievable time focused goal to work on. Although the child may have a life limiting condition the family always want a developmental goal to work towards as it helps them to focus and have a meaningful purpose to the child’s care” [ID.10].

Similarly although identified as the fifth ranked goal, “the child is cared for at home” did not achieve consensus in the third round (IQR = 3.5). Exploration of the textual commentaries provided by panelists suggests that while in many cases this is an important goal, in others it is neither desirable nor achievable. One panelist commented

“I have had a number of families who did not wish the child to be cared for at home, so while this is true in many cases it is not always so” [ID. 06],

The two lowest ranking goals, “the child’s life is prolonged” and “the family is provided with the hope that things will get better”, retained their position between rounds two and three. Both achieved consensus in the third round suggesting agreement amongst the panel that

these were not priority goals in the care of young children with life-limiting neurodevelopmental disabilities and their families.

The IQR for eleven of the thirteen goals of care were reduced between rounds two and three demonstrating a move towards consensus between the rounds. This did not hold true however for two goals, namely “parents are supported with the provision of care” and “achievement of a seamless web of care” which demonstrated a small increase in IRQ of .25 and 1.25 respectively between rounds.

Wilcoxon’s Signed Rank Test indicated stability of responses between rounds two and three for twelve of the thirteen goals with $p \leq 0.05$ set as the significance level [Table 7.4]. The only goal not to achieve response stability was “the family continues to function as a unit and enjoy life” ($W = -2.03, p = .04$).

7.7.4.2 Expert Opinion on Current Services

This section presented the findings related to the expert panel’s agreement with the service descriptor statements. It is presented according to the coding framework categories previously described, which include: the structure and funding of services; services available and the process of delivery; service integration and coordination; access to services; services’ relationship with parents; and issues associated with the provision of palliative care. Responses to the service descriptor statements are presented in Tables 7.5 to 7.10. Consensus ($IQR \leq 1$) was reached on 15 service descriptor statements in Round Two (44%), and on an additional eight service descriptor statements in Round Three, bringing the total number of statements on which consensus was reached in the study to twenty two (65%).

7.7.4.2.1 The Structure and Funding of Services

Group agreement, and consensus, was achieved on all statements related to the structure and funding of current services to young children with life-limiting neurodevelopmental disabilities and their families. The expert opinion of this Delphi panel was that current services are insufficiently funded and under-resourced to meet the needs of this population of children and their families, which results in the need to ration services to the population. The

panel agreed that current services are geographically inequitable, with a heavy reliance upon charity services to meet the needs of these children and their families. Wilcoxon's Signed Rank Test indicated stability of responses between rounds for all items [Table 7.5].

Table 7.5 *The Structure and Funding of Services*

R2 Mean	R2 Median	R2 IQR	n	R3 Mean	R3 Median	R3 IQR	n	Agreement	W	p
Children with life-limiting neurodevelopmental disabilities and their families are well served by current services										
2.00	2.00	0.75	12	2.00	2.00	0.00	12	83% disagree	-1.00	.32
There is a heavy reliance upon charity services to meet the needs of this population										
4.67	5.00	1.00	12	4.58	5.00	1.00	12	100% agree	0.00	1.00
The services available to the child in the community are dependent upon where the child lives.										
4.46	5.00	1.00	13	4.50	5.00	1.00	12	92% agree	-1.00	.32
Health services are under-resourced with respect to the services required for these children and their families										
4.69	5.00	0.50	13	4.92	5.00	0.00	12	100% agree	-1.00	.32
Insufficient funding results in the need to ration services to these children and their families										
4.15	4.00	1.50	13	4.42	4.00	1.00	12	100% agree	-1.63	.10

Shaded statements indicate agreement & consensus achieved in Round Three
Scale: 1 = strongly disagree – 5 = strongly agree

7.7.4.2.2 Services Available and the Process of Delivery

Of the ten service descriptors related to the availability and delivery of services to young children with life-limiting neurodevelopmental disabilities and their families, expert agreement was reached on six statements, and group consensus on eight statements [Table 7.6]. Wilcoxon's Signed Rank Test indicated stability of responses between rounds two and three for all items.

The expert panel agreed on specific deficits in current services provided to this group of children and their families. These included: a lack of key workers, insufficient respite services, and the provision of insufficient psychological support to both the parents and siblings of children with life-limiting neurodevelopmental disabilities. Home care was also agreed to be problematic, with the panel agreeing that children and families suffer because of long delays in obtaining necessary equipment, and disagreeing that there is good home

support for end-of-life care. Four statements did not reach the threshold for agreement set in the study.

Table 7.6 *Services Available and the Process of Delivery*

R2 Mean	R2 Median	R2 IQR	n	R3 Mean	R3 Median	R3 IQR	n	Agreement	W	p
A lack of key-workers for families results in the ad hoc delivery of services to this population										
3.83	4.00	0.75	12	4.08	4.00	0.00	12	92% agree	0.00	1.00
There is sufficient psychological support available to parents of children with life-limiting neurodevelopmental disabilities										
1.75	1.50	1.00	12	1.50	1.50	1.00	12	100% disagree	-1.00	.32
Current respite services are sufficient to meet the needs of families										
2.08	2.00	1.00	12	2.08	2.00	1.00	12	83% disagree	0.00	1.00
There is sufficient psychological support available to siblings of children with life-limiting neurodevelopmental disabilities										
1.58	1.00	1.00	12	1.50	1.00	1.00	12	92% disagree	-1.00	.32
There is good home support for end-of-life care for these children										
2.08	2.00	1.75	12	1.75	2.00	1.00	12	92% disagree	-1.00	.32
Children and families suffer because of long delays in waiting for necessary equipment										
4.15	4.00	1.00	13	4.08	4.00	1.00	12	83% agree	0.00	1.00
Services are flexible enough to respond to a family crisis at short notice										
2.25	2.00	2.75	12	2.50	2.00	1.75	12	No agreement	-1.00	.32
Out patient appointments are prioritised in favour of these children and their families										
2.17	2.00	1.75	12	2.33	2.00	1.00	12	No agreement	-1.00	.32
The lack of staff within services results in children not receiving the care they need										
4.00	4.00	1.50	13	3.83	4.00	0.75	12	No agreement	-0.58	.56
It is difficult to get a holistic view of the child's needs because of the number of health-professionals and services involved in the care of the child										
3.38	4.00	2.00	13	3.42	4.00	1.75	12	No agreement	0.00	1.00

Shaded statements indicate agreement & consensus achieved in Round Three
Scale: 1 = strongly disagree – 5 = strongly agree

7.7.4.2.3 Service Integration and Coordination

Of the six service descriptors related to the integration and coordination of services expert agreement was reached on only two statements, and group consensus on a third [Table 7.7]. Wilcoxon's Signed Rank Test indicated stability of responses between rounds for all items.

Table 7.7 Service Integration and Coordination

R2 Mean	R2 Median	R2 IQR	n	R3 Mean	R3 Median	R3 IQR	n	Agreement	W	p
There is poor communication between acute services and community based services in the care of these children										
4.00	4.50	1.75	12	4.08	4.00	1.00	12	83% agree	-1.41	0.16
Acute services are not aware of the range of services that are available to children and their families in the community										
3.54	4.00	1.00	13	4.00	4.00	0.00	12	92% agree	-1.34	0.18
General Practitioners lack the experience and expertise necessary to deal effectively with these children										
3.92	4.00	2.00	12	4.08	4.00	1.75	12	No agreement	-.100	0.32
There is poor coordination and integration of services involved in the care of these children and their families										
4.08	4.50	1.75	12	4.00	4.00	1.75	12	No agreement	-0.82	0.41
Families receive conflicting information about their child from different services										
3.26	4.00	1.00	13	3.58	4.00	1.00	12	No agreement	-1.00	0.32
There is collaboration between the different services regarding the goals of care for these children										
2.54	2.00	3.00	13	2.00	2.00	2.00	11	No agreement	-1.34	0.18

Shaded statements indicate agreement & consensus achieved in Round Three
1 = strongly disagree – 5 = strongly agree

The group was in agreement that there are some difficulties relative to the interface between acute and community based services, specifically that there is poor communication between these two levels of care. The panel also agreed that acute services are not aware of the range of services available to children and their families in the community. No other statement reached the study's threshold for agreement.

7.7.4.2.4 Ease of Access to Services

Agreement and consensus was achieved with regard to both statements in this category, and Wilcoxon's Signed Rank Test indicated stability of responses between rounds [Table 7.8]. Expert opinion was unanimous, with 100% of respondents disagreeing that it is easy for parents to access information about the services that are available for their child, or to access the services themselves.

Table 7.8 *Ease of Access to Services*

R2 Mean	R2 Median	R2 IQR	n	R3 Mean	R3 Median	R3 IQR	n	Agreement	W	p
It is easy for parents to access the services their child needs										
1.85	2.00	1.00	13	1.75	2.00	0.75	12	100% disagree	-1.00	.32
It is easy for parents to get information about the services that are available to their child										
2.00	2.00	0.00	13	1.92	2.00	0.00	12	100% disagree	-1.00	.32

Shaded statements indicate agreement & consensus achieved in Round Three

1 = strongly disagree – 5 = strongly agree

7.7.4.2.5 Perceptions of Parents

The panel failed to agree on any of the three services descriptors related to services perceptions of parents, with Wilcoxon's Signed Rank Test indicated stability of responses between rounds for all items suggesting no agreement amongst the group in respect of the position parents' occupy in the care of the child [Table 7.9].

Table 7.9 *Perceptions of Parents*

R2 Mean	R2 Median	R2 IQR	n	R3 Mean	R3 Median	R3 IQR	n	Agreement	W	p
The child and family are seen as a single unit of care										
2.67	2.50	1.00	12	2.33	2.00	0.75	12	No agreement	-1.34	0.18
Health professionals acknowledge parents expertise in the care of their child										
3.46	4.00	2.00	13	3.42	4.00	1.75	12	No agreement	-0.45	0.66
Parents are considered equal partners in the setting and prioritising of goals for their child's care										
3.46	4.00	1.50	13	3.50	4.00	1.00	12	No agreement	-1.00	0.32

1 = strongly disagree – 5 = strongly agree

7.7.4.2.6 Provision of Palliative Care

There was little agreement amongst the panel with regards to the current provision of palliative care to young children with life-limiting neurodevelopmental disabilities and their families. Of the six statements related to this aspect of services only one achieved both agreement and consensus in the final round. Specifically the expert panel agreed that in current services palliative care is considered only late in the child's condition or in cases of

crisis management. Wilcoxon Signed Ranks Test indicated stability of responses with no statistically significant change to scores between rounds two and three [Table 7.10].

Table 7.10 *Expert Opinion on the Provision of Palliative Care*

R2 Mean	R2 Median	R2 IQR	n	R3 Mean	R3 Median	R3 IQR	n	Agreement	W	p
Palliative care is only considered late in the child's condition or in crisis management										
4.50	4.50	1.00	12	4.25	4.00	1.00	12	92% agree	0.00	1.00
Medical teams lack interest in these children because of their limited prognosis										
2.75	2.00	2.75	12	4.42	2.00	1.75	12	No agreement	0.00	1.00
Children with life-limiting neurodevelopmental disabilities often undergo futile investigations and procedures										
3.50	3.50	2.50	12	3.42	3.50	1.00	12	No agreement	0.00	1.00
Medical staff are reluctant to discuss the fact that children are "life-limited" with parents										
3.92	4.00	1.75	12	3.67	4.00	2.75	12	No agreement	0.00	1.00
Medical teams fail to recognise the palliative care needs of the child										
3.85	4.00	2.50	13	3.83	4.00	2.00	12	No agreement	-1.00	0.32
Access to specialist palliative care services is readily available if it is required										
2.46	2.00	3.00	13	2.42	2.00	1.50	12	No agreement	-1.41	0.16

Shaded statements indicate agreement & consensus achieved in Round Three
1 = strongly disagree – 5 = strongly agree

7.7.4.3 Priorities for Improving Services

In Round Two panellists identified the need for a single care plan for use across services, a key worker available to every family, and a greater level of communication between all of the health professionals involved in the care of the child as the highest priorities for improving services to children with life-limiting neurodevelopmental disabilities and their families. However, the large interquartile ranges for all items in this round demonstrate considerable disagreement amongst the group with regards to the priority awarded to these changes. Only one low ranking priority (a national directory of services) achieved consensus in Round Two [Table 7.11].

Six priorities for improving services retained their original ranking between Rounds Two and Three, with an additional ten moving up or down one ranking between the rounds.

Wilcoxon's Signed Rank Test demonstrated that there was no significant difference in the mean scores for the priorities for improving services between rounds ($W = .00$, $p=1.00$).

While there was some movement in terms of priority awarded, the five highest ranking priorities for change from Round Two were retained as the five highest ranking priorities in final round, with three achieving consensus. These included "a single care plan for use across all services" ranked as the highest priority change, "a greater level of communication between all the health professionals involved in the care of the child" ranked second highest priority, and "a key worker available to every family" ranked third.

Although ranked fourth and fifth respectively neither "a greater level of coordination and integration of services" nor "a specialist paediatric palliative care consultant to act as a resource when necessary" achieved consensus in round three with IQRs of 3.5 and 6.25 respectively, indicating a wider range of disagreement amongst the group with regards to the final ranking of these priorities.

Table 7.11 *Respondents Rank Order of Priorities for Improving Services Rounds 2 & 3*

Shaded areas indicate group consensus

Required Service Change	R2 Priority	R2 Mean	R2 Median	R2 IQR	n	R3 Priority	R3 Mean	R3 Median	R3 IQR	n	W	P
A greater level of communication between all the health professionals involved in the care of the child	3	5.77	4.00	7.00	13	2	3.75	3.00	1.75	12	-1.47	.14
A key worker available to every family	2	4.92	3.00	8.50	13	3	3.92	2.50	1.00	12	-.96	.34
A single care plan for use across all services	1	4.38	3.00	5.00	13	1	2.75	1.00	1.00	12	-2.23	.03
A greater level of coordination & integration of the services involved in the care of the child	5	7.08	6.00	7.00	12	4	5.50	5.00	3.50	12	-.94	.34
A single point of contact for information for families	7	8.62	8.00	8.00	13	10*	9.33	9.00	6.50	12	-.38	.71
Less beurecracy with regards to the family's entitlements	13	11.00	12.00	7.50	13	14	11.83	12.50	6.25	12	-.55	.58
Access to palliative care in a timely and efficient manner	6	7.85	5.00	3.00	13	6	7.33	6.00	5.25	12	-.55	.58
Parent held medical records	18	12.92	15.00	9.50	13	17	14.83	17.50	7.75	12	-1.83	.07
A national directory of services	17	12.85	13.00	2.00	13	18	14.92	15.00	4.75	12	-2.04	.04
Improved education for community based health professionals	14	11.46	13.00	5.50	13	13	11.75	13.00	5.50	12	-.92	.36
A specialist paediatric palliative care consultant to act as a resource when required	4	6.46	5.00	4.00	13	5	7.08	5.50	6.25	12	-.68	.50
A formal care coordinator in every HSE area	11	9.54	10.00	9.00	13	11	10.75	11	6.50	12	-.68	.50
Medical priority status in A& E and OPD departments	12	9.77	9.00	8.50	13	12	10.92	12.00	8.25	12	-1.60	.11
The development of community based paediatric palliative care teams	8	8.85	8.00	9.50	13	9*	9.33	7.50	7.25	12	-.37	.72
Improved respite facilities	9	8.69	7.00	7.50	13	8	9.17	9.50	4.00	12	-.18	.85
A less protracted system for ordering essential equipment	16	12.38	12.00	9.00	13	16	12.17	12.50	6.75	12	-.32	.76
Improved communication between acute and community services	10	9.23	9.00	10.00	13	7	8.58	9.00	7.00	12	-.68	.47
National standards of care	15	11.54	13.00	8.50	13	15	12.00	15.50	9.50	12	-2.03	.04

* Same mean score recorded. Items ranked using median score.

In addition to those items on which consensus was achieved, the IRQ was reduced for a further 13 items in Round Three suggesting a move towards consensus in this round. However, overall the range of IQR remained wide (3.5 – 9.5) which suggests that, excluding those items which were ranked as being the three most important priority changes to services, and on which consensus was reached, there was relatively little agreement amongst the panel as to the priority service changes should take to improve the care provided to children with life-limiting neurodevelopmental disabilities and their family.

7.8 Summary of Key Findings from Phase Two

The expert panel in this Delphi identified the most important goals of care for young children with life-limiting neurodevelopmental disabilities and their family as:

1. Achieving the best possible quality of life for the child.
2. Open and honest communication with the family.
3. Achievement of the child's full potential within the limits of the illness.
4. Optimum symptom management.
5. The child is cared for at home.

There was a high level of agreement amongst the expert panel that these should be the primary goals when providing care to this population of children and their families.

At a general level there was agreement about issues related to the structure and funding of services to young children with life-limiting neurodevelopmental disabilities and their families. The panel agreed that current services do not well serve this population, with under-funded and under-resourced services, and a heavy reliance upon charity services to meet the needs of this group of children and their families. They also agreed that the availability and provision of services is largely dependent upon the geographical region in which the family lives.

The panel also agreed that there are gaps in some service areas, as well as a lack of communication between acute and primary care services. Specific service deficits included; insufficient respite services; insufficient psychological support to parents and siblings; poor home support for end-of-life care; long delays in obtaining necessary equipment; and a lack

of key workers available to children and their families. Access to both services, and information about services, was agreed to be difficult for parents.

The position that parents occupy in the context of the services their child received is not clear, with little agreement amongst the expert panel in this regard. Similarly the provision of palliative care to this population of children appears complex, with little agreement amongst the expert panel beyond agreeing that palliative care is currently only considered late in the child's condition or in crisis management.

The panel identified what they considered to be the most important changes to services that would act to improve the care delivered to children with life-limiting neurodevelopmental disabilities and their families. These were identified as

1. A single care plan for use across all services
2. A greater level of communication between all the health professionals involved in the care of the child
3. A key worker available to every family
4. A greater level of coordination and integration of the services involved in the care of the child
5. A specialist paediatric palliative care consultant to act as a resource when required.

However whilst there was a high level of agreement amongst the panel with regards to the top three requirements for improving services to young children with life-limiting neurodevelopmental disabilities and their families, the large IQR for the fourth and fifth indicated much less agreement regarding these particular priorities for change.

7.9 Preliminary Discussion

This section discusses the findings from the Delphi used in this phase of the study. Both consistencies and inconsistencies in the findings are discussed.

It is the explicit and stated aim of Irish health policy to provide health services to all who need them. Indeed the vision stated by the Department of Health and Children is for “a health services that is there when you need it, that is fair, and that you can trust” (DOH&C, 2001b:8), while the children's palliative care policy (DOH&C, 2010:25) proposes the

development of children's palliative care services that are based upon the principles of "inclusiveness, partnership, comprehensiveness and flexibility". Despite these stated visions and aspirations the findings of this Delphi suggest that this has yet to be achieved, and that developing systems of services to best meet the needs of young children with life-limiting neurodevelopmental disabilities and their families continues to represent a significant challenge for all services and agencies involved in the care of this population.

The goals of care agreed by the panel for children with life-limiting neurodevelopmental disabilities and their families are generally consistent with the concept of a child-centred service. However, absent from this priority list of goals is the family-centred element of care synonymous with a palliative care approach (Sepulveda *et al*, 2002; DOH&C, 2005, 2010). It is interesting that, despite the emphasis awarded to supporting parents with the provision of care in the preliminary interviews, and the identification of "parents are supported with the provision of care" in the first Delphi round, this issue did not rank in the top five goals of care in the iterative rounds of this Delphi. This is despite the fact that "the child is cared for at home" did. In the context parents' reports from Phase One, this may suggest that service providers underestimate either the work involved in caring for these children at home, or the negative impact providing care has on the family especially the mother as the main provider of day-to-day care. This may account for the relatively low priority awarded by the panel to "the provision of appropriate respite" for families, and to the goal of "the family continues to function as a unit and enjoy life". It may also account for the fact that the panel did not agree that children and families are seen as a unit of care.

This apparent lack of a family-centered approach to care is reinforced by the lack of agreement amongst the panel with regards to services general perceptions of parents, whereby the panel failed to agree that parents' expertise is acknowledged in the care of their child, or that parents are considered equal partners in the setting and prioritising of goals for the child's care. Partnership is important to parents of children with disabilities (Case, 2000; Farrell *et al*, 2004, Campbell, 2007). Partnering between families and their children's' health-care providers is a cornerstone of family-centred care and has been found to be associated with demonstrably better outcomes than those achieved through the traditional medical model (Zimmerman & Dabelko, 2007; Knapp *et al*, 2010). Although there is no single definition of partnership in this context Fereday *et al* (2010) defines equality, mutual respect, mutually

agreed upon goals and shared planning and decision making amongst the constituents of a parent-professional partnership. However it may be that the idea of “partnership” is interpreted differently between services and parents. The findings of this Delphi suggest that that professionals continue to control the parent–professional relationship, assuming the role of ‘expert’, rather than integrating and consulting parents in a negotiated decision-making process. This is contrast to MacKean *et al*’s (2005) findings which suggests that parents want to work truly collaboratively with health-care providers in making treatment decisions and on implementing a dynamic care plan that will work best for child and family. Recognition and acknowledgement of parents’ expertise, not just their technical skill, is important and has been found to underpin parent-professional relationships. Kirk and Glendinning (2002) found that experts who failed to acknowledge parents expertise or who questioned parents judgment about the child’s need for treatment caused parents anger and distress, not least because of the potentially serious consequences for the child, while Knapp *et al* (2010) found that family-provider partnership was a positive and significant factor for organised and easily accessed community services.

There was agreement amongst the expert panel that services are under-resourced, under-funded, geographically inequitable and heavily reliant upon charity services to meet the needs of children with life-limiting neurodevelopmental disabilities and their families. These findings are consistent with parents’ reports form Phase One, and with previous Irish research in this area, from both the perspectives of mothers (Redmond & Richardson, 2003) and service providers (Quinn *et al*, 2005). Quinn *et al* (2005) reported that service providers in their study reported services that are difficult, inconsistent and at times unfair and discriminatory. Specific deficits in service provision identified by the Delphi panel in this study included psychological support, provision of respite, difficulty accessing necessary equipment and appliances, and in-home support for end-of-life care. In addition the panel agreed that a lack of key workers is problematic in current services. Many of these factors were also identified as inadequacies by health service providers in Quinn *et al*’s study (2005). Psychological support for parents of young children with life-limiting neurodevelopmental disabilities is essential since a significant body of literature indicates that parents of children with disabilities experience significant stress which may be chronic and persistent over time (Hodapp *et al*, 2003; Redmond & Richardson, 2003; Forde *et al*, 2004; Raina *et al*, 2005; Estes *et al*, 2009). Since respite has been demonstrated to ameliorate psychological distress in

parents of children with neurodevelopmental disabilities (Mullins *et al*, 2002) it is possible that the lack of both respite services and psychological support acts synergistically to increase the overall burden on mothers.

Despite the fact that key workers are a key feature of current policy initiatives (DOH&C, 2005; DOH&C, 2010), and have been demonstrated to have positive outcomes for families (Greco & Sloper, 2004; Greco *et al*, 2005; Sloper *et al*, 2006), the panel agreed that the lack of key workers continues to be problem in current services to children and their families. This issue was also raised by parents in Phase One.

There also appeared to be some contradictions and inconsistencies in the findings of this Delphi. Gibson and Brown (2009:141) suggest that inconsistency occurs when “two or more features of the data are not consistent with each other”. This is apparent in the context of the expert panel’s opinion on the integration and coordination of current services. There are two main issues to be explored here. The first relates to the dichotomy between the opinion of this expert panel and the expressed opinion of parents in Phase One and previous research carried out in an Irish context by Redmond and Richardson (2003) and Quinn *et al* (2005). The second relates to the inconsistency between the panels opinion on service coordination and the proposal of “improved service coordination and integration” as the fourth priority change that would improve services to this population of children and their families.

Expert opinion in this Delphi appears to not only contradict previous research in this area (Redmond & Richardson, 2003; Quinn *et al*, 2005), and also contradicts parents’ experiences of service in Round One whereby parents reported fragmentation in the way services are organised and delivered. The inconsistency between the opinion of this panel and the opinion of health professionals in Quinn *et al*’s (2005) study may possibly be accounted for by methodological variations in the two studies. Quinn *et al* (2005) used a qualitative approach to explore professionals’ opinions about service integration and coordination for a generic population of life-limited children in the context of specialist palliative care. Alternatively this study used a quantitative approach to explore professionals’ opinions about general service integration and coordination specifically in the context of young children with life-limiting neurodevelopmental disability.

However, accounting for the difference in opinion between the expert panel of service providers and parents is more difficult. Due to the variety of services and agencies involved in the care of these children and their families it is possible that health professionals are focused predominantly on their own service and area of expertise, and are simply not aware of how other services function and how services fit together. This broader view may only be available to parents as they try to access and integrate services into a meaningful whole to apply them to the needs of the child and family. This position is supported by Hunt, Elston & Galloway (2003) who also reported discrepant views between parents and service providers in their study exploring service provision to life-limited children and their families in the United Kingdom. Kirk and Glendinning (2002) reported similar findings whereby professional groups feel they worked well together contrary to parents' experiences and suggests that this is related more to their valuing each other's professional contributions.

The second issue in this regard relates to the panel's proposal of "improved service coordination and integration" as the fourth priority change that would improve services to this population of children and their families, despite the fact that they failed to reach agreement that there is poor coordination of current services. The discrepancy may be a methodological issue associated with the varied locations and small number of experts on the panel. It is also possible that poor coordination of services is not a national problem and that service integration and coordination is worse in some areas than in others which would contribute to this discrepant view.

There was little agreement by the expert panel on issues related to the provision of palliative care, or adoption of a palliative care approach, to children with life-limiting neurodevelopmental disabilities and their families. The expert panel agreed that palliative care is only considered late in the child's condition or in cases of crisis management, but did not agree that medical teams fail to recognise the palliative care needs of the child, or that medical teams are reluctant to discuss that the child is "life-limited" with parents. There are a number of possible explanations for this apparent contradiction. Based upon parents reports in Phase One of the study (parents perceptions that generally their child's symptoms are adequately controlled) it may be that early referral is not necessarily indicated or required in this group of children. Alternatively, since the panel did not agree that access to specialist palliative care services is readily available if needed, it may be an issue of access. This would

account for the identification of a specialist paediatric palliative care consultant as a priority for service improvement by panellists. It is also possible that late referral is a consequence of the difficulty of defining the terminal phase of illness in this group of children (Levetown, 2001; Brook & Hain, 2008) which is potentially problematic for this group of children and their families in the context of the DOH&C children's palliative care policy (2010) which advocates that priority for in-home support is given to children who are "approaching the end of life".

The top three priority changes to improve current services are consistent with the expert panel's agreement that there is poor communication between hospital and community based services, and this supports parents experiences from Phase One. The centrality of a coordinated and multiagency approach to the planning and delivery of care and support to all children with palliative care needs has been documented (Watson *et al*, 2002b; ACT, 2003; Quinn *et al*, 2005; DOH&C, 2010). Campbell (2007) identifies that, in the context of disability services, the highest quality ratings are achieved when there is evidence of the use of multidisciplinary integrated care pathways which clarify expected steps and outcomes. Similarly the priority awarded to a key-worker for every family is consistent with the panel's agreement that the current lack of this resource results in the ad hoc deliver of services. This is consistent with finding of pervious research which suggest that key-worker availability would improve coordination and integration of services to the child and family Hunt, Elston & Galloway, 2003; Quinn *et al*, 2005; Greco *et al*, 2005; Sloper *et al*, 2006).

The second inconsistency in the data relates to the panels proposal for the appointment of a specialist palliative care consultant as the fifth priority change to services that would improve care to this population of children and their families. This appears inconsistent with the panel's opinion on current service provision especially the failure to agree on issues of access to specialist palliative care. One explanation for this may be the difference between having access to an adult specialist palliative care service adapted to the needs of a child with a life-limiting neurodevelopmental disability as is currently available, and access to a specialist paediatric palliative care services, which would be focused exclusively on the needs of the child and family, as is the proposed change to current services.

Although the top five priorities for service improvement were consistent between Rounds Two and Three, the IQR for proposed changes to improve services was much larger than the IQR for the goals of care indicating less agreement amongst the panel with regard to the factors needed to improve services. This may be a function of the panel constituency in that members were both hospital and community based, and it is possible that the perceptions of these two groups may differ with regards to the potential of changes required to improve services. An alternative explanation may be that, although professionals are generally agreed on the goals of care for these children and their families, they are simply less likely to agree on how services should function to achieve these goals.

7.10 Strengths and Limitations of Phase Two

The conclusion drawn from this phase of the study should be considered in the context of the strengths and limitations of the phase. There is little definitive guidance to be found in the literature with regards to the decisions made during a Delphi, and although the decisions made, and the rationale for these decisions, have been made clear in this study, it is nevertheless subject to the general criticisms leveled at the Delphi method.

The main strength of this Delphi was the nature and constituency of the expert panel. The criteria for expertise are clearly stated, and the panel encompassed a comprehensive, interdisciplinary, multi-agency perspective. All services, statutory, voluntary and charity services which provide care to this population of children and their families were included. Many professional disciplines were involved and all levels of services delivery were represented. In addition, the panel represented a national sample of experts.

Alternatively the overall size of the panel was relatively small when compared to the panel size in much published literature, and confidence in the findings needs to be considered in the context of the panel size. It is difficult to directly compare the response rate for this Delphi with other published literature due to the variety of methods used for reporting response rates and, in some cases, the ambiguity of exactly what is being reported. Some authors report an overall response rate, for example Gibson (1998) reported a 64% response rate in a three round Delphi, while Butterworth and Bishop (1995) report a response rate of 61% in their two-round Delphi, however it is not clear whether this figure represents the figure for each

round, or the percentage of respondents who completed all rounds. Although it is generally accepted that the higher the response rate the greater the validity of the study, there is little guidance in the literature regarding a definitive standard. Sumsion (1998) suggests a minimum response rate of 70% for each round although this appears to be an arbitrary figure and the empirical justification for it as a significant standard is not clear. Although some studies do report response rates in excess of 90% for each round (Crouch *et al*, 2002; Staggers *et al*, 2002; Hemmings *et al*, 2009) more commonly this 70% minimum response rate for each round is not achieved with a minimum range of 40 – 65% reported in the literature (Endacott *et al*, 1999; Scott *et al*, 2006; Wiener *et al*, 2009; Green *et al*, 2009; Wilson & Moffat, 2010). This study met Sumsion's (1998) standard for all but the first round.

7.11 Conclusion

The expert opinion of this Delphi panel is that currently services to young children with life-limiting neurodevelopmental disabilities and their families are under-funded and under-resourced, with definite gaps in some areas of service provision as well as poor communication between acute and community based services. While the expert panel agrees on what the goals of care for this population of children and their family are, there is less consensus regarding the changes to current services that are required to achieve these goals and improve services to this population of children and their families. This makes acting to improve services to young children with life-limiting neurodevelopmental disability a complex task.

This chapter has provided an overview of Delphi and described the conduct of a Delphi study of expert opinion on the services currently available to young children with life-limiting neurodevelopmental disabilities and their families. Both agreement and disagreement amongst the panel had been explored and possible explanations of inconsistencies in the findings proposed. The findings from this Delphi will be integrated with those from Phase One of the study in the next chapter.

Chapter 8: Integration of Findings

Segal's Law

“A man with one watch knows what time it is
A man with two watches is never sure”

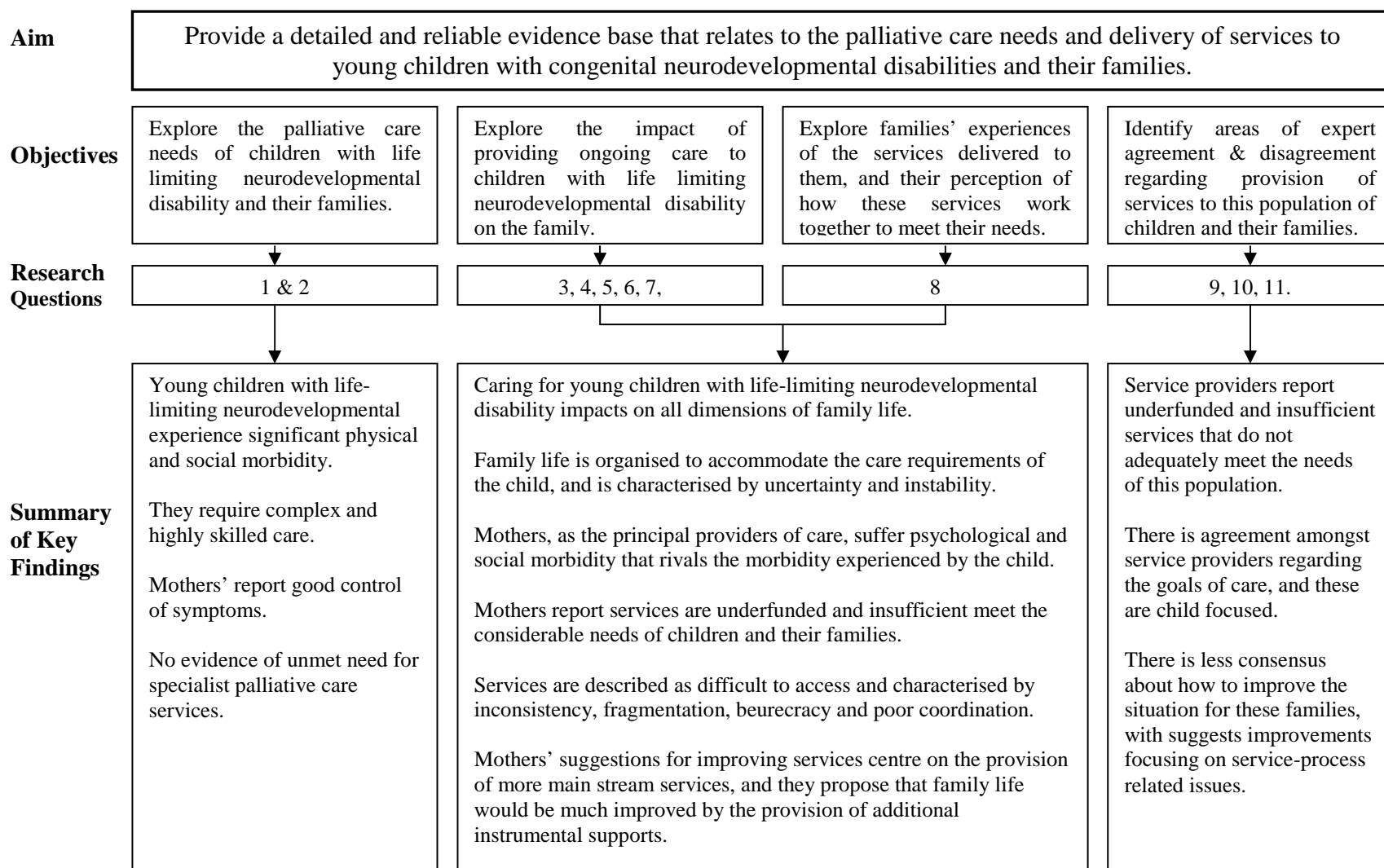
Bloch A. (2003:36) Murphy's Law.
New York; Perigee Books

8.1 Introduction

In this chapter the integration of findings from all phases and stages (parents' survey, mothers' interviews and Delphi) are presented. This presentation is sequenced in accordance with the research objectives and questions identified in Chapter Four. Subsequently the chapter begins with the integrated findings related to the challenges experience by young children with life-limiting neurodevelopmental disabilities and their palliative care needs. These findings address research questions one and two. This is followed by the findings related to the impact of providing care on the family. This focuses principally upon the impact on, and perspectives of, the mothers of young children with life-limiting neurodevelopmental disabilities as mothers were the principal respondents in both stages of this study and also identified themselves as the main providers of care to the child. These findings relate to research questions three and four and also questions six to eight. Finally the integrated findings related to the services delivered to this population of children and their families are presented. These findings relate to research question five and questions nine to eleven.

An abbreviated version of the findings relating these to the aims, objectives and research questions of the study are presented in Figure 8.1.

Figure 8.1 *Integration of Findings: Meeting the Aims and Objectives of the Study*



8.2 Young Children with Life-Limiting Neurodevelopmental Disabilities and Their Palliative Care Needs

Data from parents suggest that young children with life-limiting neurodevelopmental disability experience significant physical and social morbidity associated with their conditions. Often a wide range of medical intervention and technology was required to sustain the child on a day-to-day basis. While individuals problems or particular symptoms experienced by the child were generally considered to be well controlled, overall the management and care of these children involved considerable labour on the part of mothers, who were required to provide high levels of often complex and technologically advanced care on a daily basis. Not only did this care involve the management of complex drug regimes and feeding schedules, but it also required mothers to develop specific skills in areas of nursing and physiotherapy. Mothers' reported that they received little or no training for this aspect of care.

In many cases the care required by the child involved round-the-clock, twenty-four hour a day care. The provision of this care was characterised by uncertainty, unpredictability, and frequent oscillation in the child's health status. This was reported in the survey's open-ended questions, whereby mothers commented on the complexity and instability of the child's condition, and described in detail in mothers narratives. The unpredictable nature of the child's condition meant that mothers needed to be constantly vigilant and alert to even the most subtle changes in the child's normal health status, it also made obtaining outside help with the child's care difficult. In an attempt to maintain stability in the child's condition, and to ensure the very best for their children, mothers established a strict and precise routine round compliance with the child's medical and technological needs. This routine took precedence over all other aspects of family life, and all other members of the family unit.

In addition to the physical and medical challenges confronting the child, mothers' also described the considerable social morbidity that the child experienced as a consequence of their condition. This involved the exclusion of the child from the everyday social opportunities normally available to young children.

Despite all of these challenges, and the significant and unrelenting ordeal of care-work involved, children in this study were much loved and valued. This was an essential feature of mothers' narratives. Although mothers described the difficulties involved in providing ongoing care, and many reported a need for greater respite facilities and other practical supports to help them provide the care required, at no juncture in the study did any mother suggest that they wished for their child to be cared for anywhere but in their own home.

8.3 The Family Impact of Providing Ongoing Care

The consequences of providing on-going care to young children with life-limiting neurodevelopmental disabilities are significant, for the family unit, and in particular for the mother as the main provider of care.

Mothers in this study experienced significant psychological morbidity. This is evidenced in the total GHQ-28 scores amongst the sample. The high scores on the somatic symptoms and social dysfunction subscale of the GHQ-28 is further explained in mothers' stories of physical burden and exhaustion, and the curtailment of opportunities for social interaction outside the home as a consequence of the care work required. Some mothers lamented the loss of employment, both as an issue of changed self-concept and as an opportunity for social interaction, while others spoke of the difficulties of maintaining friendships in the context of complex and unpredictable care obligations. The Anxiety and Insomnia subscale of the GHQ28 recorded the highest mean subscale score amongst the mothers in this sample. Mothers' interviews would suggest that most of this anxiety is associated with service-related issues, in particular the constant apprehension about continued and appropriate service provision to their child, and for many, the worry about how they would continue to provide care in the future should services be reduced or withdrawn. The insomnia element of the subscale may also be influenced by the 24hr nature of the work involved in caring for a young child with a life-limiting neurodevelopmental disability, and the unpredictable nature of the child's condition. The high level of psychological morbidity recorded in Stage One of the first phase is consistent with the expressed view of the expert panel that there is not enough psychological support available to parents.

Although mothers recorded fairly good levels of social support in Stage One, the importance of distinguishing between social support as a general concept and practical help as a specific concept became apparent in Stage Two. It is evident from mothers' narrative reports, and from the commentary that mothers made in the open-ended section of the social support scale used in Stage One, that although partners, family and friends are generally supportive this does not necessarily translate into practical assistance that is of great value in the day-to-day provision of care. Mothers considered that friends had their own lives to get on with or did not understand the complexity of their situation, and often did not want to worry other family members with their concerns and difficulties. In addition, because of the complex nature of the child's care, a standard of technical or medical proficiency was required which was often not available from family and friends.

Providing on-going care to a young child with a life-limiting neurodevelopmental disability at home also had consequences for the family unit. This was evidenced by the IFS scores in the parents' survey, and explained in mothers' interviews. This negative impact appeared to be subtle and multi-faceted, and the predictor variables for this were not easy to identify. Mothers' described that family relationships were impacted. This appeared to be particularly problematic if there were other siblings in the family. In both stages of the study mother reported the time commitment required to meet the needs of the ill child negatively affected the amount of time available to devote to the needs of other siblings. This was a cause of considerable guilt in some instances. However mothers also reported an impact on relationships with partners as the changed family circumstance necessitated a re-negotiation of roles and relationships.

There were also consequences for family finances, with mothers' reporting increased costs associated with frequent hospitalisations, additional medications not available under the General Medical Services Card scheme, travel to appointments, and equipment that they considered essential to their child's needs but which was not available to them through services.

Mothers coped day-to-day in a variety of ways. They drew upon several sources of support including partners, family, friends and the parents of other children in situations similar to their own. Coping was facilitated by adhering to a strict routine for the child's care,

organising family life with military precision, focusing on and accentuating positive elements, and by attributing meaning to the situation. Coping appeared to have a bidirectional focus and was aimed at achieving two main goals: ensuring the needs of all family members were met, and maintaining a sense of normalcy for the child and the family.

8.4 The Services Available and the Process of Delivery

While there was a high level of service usage amongst the families in this study there was also considerable unmet need for services. This did not appear to be related to a requirement for specialist services, rather it was associated with the need for more frequent access to the general services that the child currently received. In particular, mothers expressed an urgent need for more therapeutic services and family support services. Mothers' experiences and perceptions of the services available to their child and family were consistent between the first and second stages of Phase One. Survey findings that services are insufficient, inconsistent, and underfunded received strong support from mothers' narratives. In addition, these perceptions were supported by the opinions of service providers in Phase Two, whereby the expert panel also expressed that current services are underfunded, under-resourced and insufficient to meet the needs of this population of children and their families. Both groups of respondents reported that they believed there was an inequitable geographical distribution of resources, with services related not to the level of need experienced by the child and family, but rather dependent upon where the family lived.

Given this uniform perception of the services currently available it is not surprising that both parents and service providers reported a heavy reliance on charity services to meet the needs of this population of children and their families. This was identified in parents' surveys and amply evidenced in mothers' narratives where they reported not only their appreciation of, and dependence on, a particular charity service, but describe this service as essential support to their continued coping. Several mothers reported that the potential loss of this service was a source of constant fear and anxiety for them.

Mothers also described additional difficulties they encountered with the services that their child receives which were acknowledged by service providers. These included difficulties with accessing information about services, long delays and waiting times, and cumbersome

bureaucracy that substantially increased the overall burden of care. Both mothers and service providers acknowledged the difficulties encountered while trying to access services for the child. Accessing what their child needed was described by mothers as a constant “battle”. This metaphor was used in parents’ surveys and repeated frequently in mothers’ narratives. Mothers’ narratives suggested that it was this constant struggle with services that had more negative consequences, for the child, the mother and the family unit, than the substantial work involved in caring for the child day-to-day. While the expert panel reported poor communication between acute and community services, communication was a much more critical issue for mothers in this study who reported considerable difficulty with communication at all levels.

The panel of service providers opinion that the lack of key-workers results in the ad hoc delivery of services to young children with life-limiting neurodevelopmental disabilities and their families is consistent with parents identification of the need for a single source of support and information to improve their family situation (many specified that this was a key-worker). The issue of key-workers also arose, both directly and indirectly, in both stages of the parents’ data. Having a key-worker was negatively correlated with mean IFS scores, and with difficulty obtaining services. In addition having a key-worker or identified link-person was also identified as a factor that worked well, when it was available, in parents’ surveys. Mothers’ narratives supported the view that having a named link-person was a critical resource when it came to accessing information and services that they needed for the child, yet findings from Stage One suggest that one in four families do not have access to a key-worker or link-person.

Relationships with health care providers arose as another important, but somewhat conflicted, feature of this study. Respectful and Supportive care recorded the highest mean score of the MPOC20 subscales in Stage One, and all mothers recounted individuals who had a significant positive impact on their family situation in Stage Two. However, the Enabling and Partnership subscale recorded only a mean score of 4.6 suggesting that parents experience this only to a moderate extent, and a lack of partnership orientation on the part of services is evident in mothers’ narratives in Stage Two with mothers’ reporting that their experience and expertise in the care of their child is frequently under-valued or ignored by service providers. This perception is supported by the panel’s failure to agree on aspects of partnership with

parents, and the ambivalence of the panel relative to services perceptions of parents in general.

Despite these difficulties, mothers' perceptions of services were not routinely negative in this study. In both stages of Phase One mothers consistently identified factors that they felt worked well for their children and themselves. In the quantitative round mothers rated the relationship / attitudes of staff as the highest positive factor in current services, closely followed by a named charity service available to their child and themselves. Both of these factors were re-iterated in interviews where mothers were anxious to point out the caring, sympathetic and supportive health-professionals they had encountered. Despite the significant difficulties and challenges that mothers reported and described they rated themselves as "fairly satisfied" with the service they receive overall. While it is possible that the provision of care is sufficiently burdensome, and that mothers are so overwhelmed, and subsequently so relieved to obtain any service assistance that they are satisfied with whatever they get, mothers' stories suggest that these periods of overwhelming burden are episodic and mostly related to changes in the child's condition. It appears more likely that individual services work well once they are received; it is the process of obtaining and integrating services rather than the quality of services that is the cause of difficulty. This would account for mothers' perceptions of being satisfied with individual services but simultaneously reporting that services are difficult to access and lack integration and coordination. There is strong support for this explanation in mothers' narratives of just getting on with it.

This concept of individual services working well once they are received is supported by mothers' experiences of respite services. Respite care was identified by some parents in Stage One as a service that worked well for their child and family, and yet was also identified by an almost equal number of parents in the same stage as a service they considered necessary for improving their family situation. This suggests that respite care is an essential component of family support for families of young children with life-limiting neurodevelopmental disabilities and their families. It also suggests inequitable provision of this service, and supports the reported inconsistencies and perceived inequality of service provision to this population of children and their families. Inequitable provision of respite services may result in some families having easy access to appropriate respite for their child, while other families are unable to access the same service. This explanation is supported by parents' experiences

of home-support services in Phase One where eleven parents identified respite as a factor that works well for their child and family while eight identified it as a factor they considered necessary to improve services.

8.5 Divergent Opinions between Parents and Service Providers

There were several areas of divergence between the experiences of parents and the perceptions of service providers in the study. These related to differences of opinion between the two groups about the integration and coordination of services, and on the changes to current services required to improve the care and services families receive.

While the expert panel did not agree that there is poor coordination and integration of services this was not parents' experiences service usage. Parents' reported fragmentation at all levels of service provision: between different disciplines in the services, between acute and community based services, and between different agencies working in the community.

There were also differences of opinion between parents and expert service providers with regards to the factors necessary to improve the services delivered to children and their families. Parents' requirements were essentially pragmatic, and their suggestions were principally focused on the practical things that would make life easier for the child and the family. Their main concerns were improving the frequency and ease of access to the mainstream services that are already available, closely followed by having a single source of information and support. Accessing information was a consistent challenge for mothers in this study yet having a single source of information for parents was ranked only tenth in potential to improve services to this population of children and their families by the expert panel.

Similarly while parents' identified the need for greater in-home support and respite services to ameliorate their difficulties, improved respite facilities was ranked only ninth by the expert panel in terms of its potential to improve services, and improved in-home support was not identified at all. This is despite the fact that the panel acknowledged both of these to be deficient in current services.

These divergent views demonstrate a significant gap between the opinions of service providers and the experiences of service users. The differences suggest that service providers underestimate the fragmentation of services, and overestimate the way in which the many disparate services involved in the care of these children and their families join-up to form a cohesive system of care provision. Although parents identify many factors that are deficient or defunct in the care they receive, the factors they propose as most necessary to improve care and services are primarily concerned with resources required to alleviate the practical difficulties they encounter day-to-day. Alternatively the factors identified by service providers as most necessary to improve care and services to this population of children and their families are focused more on improving the process of service delivery rather than on the provision practical resources.

8.6 Conclusion

This chapter has presented the integrated findings from the study as a whole. Convergent and divergent views have been presented. Parents' quantitative and qualitative data are generally consistent, and while there were many areas of consistency between the opinions of parents as service users and the opinions of the expert panel of services providers there were also some divergent views. Principally these are related to two areas: the manner in which services function to deliver care to young children with life-limiting neurodevelopmental disabilities and their families; and the importance afforded to factors that need to be addressed in order to improve these services and better meet the needs of these children and their families. These findings will be discussed in detail in the following chapter.

Chapter 9: Discussion of Findings

"There is always an easy solution to every human problem--neat, plausible, and wrong."

H. L. Mencken

The Devine Afflatus 1917

9.1 Introduction

This chapter discusses the findings of this study. Congruent with the use of a mixed methods design the discussion will focus on the integrated findings rather than the individual findings from the study's separate phases. As in the previous chapter, the discussion is sequenced in accordance with the study's objectives and research questions. Consequently the chapter begins with a discussion of findings related to the young child with a life-limiting neurodevelopmental disability, their palliative care needs, and the nature of the care they require. This is followed by a discussion of the impact of providing care on families. Finally the findings related to service provision for these children and their families are discussed. The relevance of the findings to the theoretical framework used to underpin the study is discussed, and the chapter concludes with a discussion of the overall strengths and limitations of the study.

9.2 Young Children with Life-Limiting Neurodevelopmental Disabilities and their Palliative Care Needs

This section discusses the findings of this study that relate specifically to the young children in the sample and their palliative care needs.

9.2.1 Morbidity Experienced by Young Children with Life-Limiting Neurodevelopmental Disabilities

The children in this study suffered considerable morbidity associated with their condition. A wide range of physical problems were reported and many children were reliant on some form of medical technology to sustain them day-to-day. Mothers described complex medical routines that needed to be performed on a round-the-clock basis. There is relatively little

literature exploring the specific morbidity experienced by young children with life-limiting neurodevelopmental disabilities, although the symptom profile of the children in this study is similar to that described by Lenton *et al* (2001) for children with a wide range of life-limiting conditions. These similarities would appear to support a non-categorical approach to exploring the needs of children with life-limiting conditions, at least in terms of the physical morbidity associated with the children's conditions.

9.2.2 Requirement for Specialist Palliative Care Services

Despite experiencing a considerable number of individual and sometimes complex problems, mothers' reported relatively good symptom control and management in this sample of children. This finding raises questions about the consistent calls for access to specialist palliative care services for this group of children (ACT, 2003, 2009; Redmond & Richardson, 2003; Carter & Levetown, 2004; Brook & Hain, 2008). Specialist palliative care services are primarily focused on the provision of disease or treatment related symptom control (DOH&C, 2001; Sepulveda, 2002) albeit with a holistic focus. The fact that symptom control was not identified by parents as a problem in this study would suggest that specialist palliative care may in fact not be routinely indicated for this group of children. The cross-sectional nature of this study makes this a tentative finding however, and it is possible, as suggested by Carter and Levetown (2004) and Brook and Hain (2008), that oscillations in the child's condition makes the timing of palliative care difficult to establish. However many of the young children in this study had had numerous, and often prolonged, episodes of hospitalisation, and although mothers identified many difficulties associated with these, the issue of poorly controlled symptoms or unnecessary suffering did not arise even in this context. The fact that two of the children in the sample were already in receipt of specialist palliative care, one for the management of complex pain, suggests that children who are identified as needing specialist palliative care are referred to such services. The second family in receipt of specialist palliative care services in this sample were referred in the absence of complex symptoms, and could not see the relevance of this service to their child. Early referral for this mother was an additional cause of distress as she believed the service had little to offer the child and conflicted with the family's own goals of care.

Calls for specialist palliative care services for this population as a whole are based upon the assumption that the service is relevant to all children by virtue of an uncertain or limited prognosis. Indeed Carter and Levetown (2004) suggest that the obvious answer to the issue of which children need palliative care services is that children who “will die” or “are dying” need proposing that palliative care is offered when it has been acknowledged that the child’s illness is likely to ultimately cause death. Without explanation or commentary on the complexity of the symptoms experienced, Redmond and Richardson (2003) suggest that it is surprising that none of the children with complex and life-limiting disabilities in their study were referred to palliative care services. This position appears to devalue, or underestimate, the efficacy of the services that this group of children currently receive. Mothers considered their child’s symptom control to be good, and there were no calls for additional specialist services to improve the child’s care or condition. Instead mothers’ consistently called for more main stream services and better in-home support services. In this context it would appear that the vast majority of young children with life-limiting neurodevelopmental disabilities may be better served by improving current mainstream services than by developing and introducing costly additional specialist services. This may account for the apparent lack of agreement amongst the expert Delphi panel with regards to the provision of palliative care to this population of children. While the panel agreed that referrals to specialist palliative care services are generally made late in the child’s condition they did not agree that medical teams fail to recognise the palliative needs of the child.

Lenton *et al* (2004) also questions the need for palliative care for children with life-long complex needs. They suggest that while the aspirations of palliative care are to offer symptom control, emotional support and practical help to the child and family, these aspirations are really no different to those of many professionals working with such children day-to-day, proposing that addressing these needs should be part of mainstream care. This position is supported by a study by Horrocks *et al* (2002) who provide a description of 18 children referred to a newly developed specialist palliative care service for children with genetic, metabolic and degenerative diseases of a life-limiting nature and their families in the U.K. The children in the sample ranged from five months to seventeen years. The study did not assess individual problems experienced by the child however seven children were rated by their parents as being “very” or “extremely” troubled on referral to the service. Horrocks *et al* (2002) report that perceived problems were as frequently due to emotional or practical

care needs as due directly to the children's physical illness, but no further description is provided beyond this. What is most interesting about this study is the fact that overall distress rating was only slightly reduced 6 months after the intervention with five parents now rating their child as "very" or "extremely" troubled, and although information and nursing care needs appeared to be improved amongst the group other difficulties such as problems with equipment and respite needs were not improved. This would appear to suggest that these children and their families require most are better services and a more holistic model of service support addressing the impact of social and psychological stress on the family.

While it may legitimately be argued that these children and their families require a palliative approach to care in the context of a focus on quality-of-life for the individual and the family, the basic elements of this approach has been widely endorsed in the general field of intellectual disability, where increasingly the construct of family quality-of-life is being used to consider issues of family needs (Brown *et al*, 2003; Summers *et al*, 2005; Brown *et al*, 2009; Werner *et al*, 2009). Park *et al* (2003:368) defines family quality-of-life as "conditions where the family's needs are met, and family members enjoy their life together as a family and have a chance to do things that are important to them". As contrasted with individual quality of life, family quality of life addresses the impact of individual quality of life on the family, consequently the concept of family quality of life focuses on family-orientated outcomes. Poston *et al* (2003) suggests a domain structure with two parts: (1) domains with an individual orientation which occur at the individual level and represent the idiosyncratic ways in which quality of life of individual members impact on the quality of life of other members and the family as a whole; and (2) domains with a family orientation which provide a context in which individual family members live their lives collectively as a family unit. In this respect the family quality of life approach is consistent with the systems theories that underpin much of family nursing (Freidman *et al*, 2003; Rowe-Haakinen *et al*, 2010). This approach focuses simultaneously on both the child and the family. It would appear that such an approach has a lot to offer families of young children with life-limiting neurodevelopmental disabilities particularly if families receive a comprehensive, coordinated approach to care aimed to bolster the family's strengths and capacity to cope. Although this family orientation is consistent with a children's palliative care approach, the findings of this study suggest that it matters little which service provides it, so long as it is effective. What is important is that families have their needs assessed, and receive appropriate and sufficient

support so that they are able to deliver the care that their child needs, and that the agencies involved work together so that this support is delivered in a way that is meaningful for both the child and the family.

9.3.2 The Care Required by Young Children with Life-Limiting Neurodevelopmental Disabilities

The move towards primary care has meant that medical and technical work, once provided by trained health care professionals, is now provided by families at home. The impact of this health care reform on families is significant, and the shift of responsibility for care to family members has produced a spectrum of issues that demand psychological, ethical, social, financial and policy solutions (Schachter & Holland, 1995). McKeever and Millar (2004) suggests that contemporary community health and social care provision requires mothers of children with chronic illnesses or disability to adopt, and adapt to, an unfamiliar and relatively low-status position of skilled, unpaid paraprofessional in fields in which most mothers engage only intermittently. Dalley (1996) suggests that mothers who provide such labour intensive and medically sophisticated care as part of their normal mothering form part of a reserve army of care providers who are unremunerated and under-acknowledged.

In terms of the nature of the work involved in caring for a young child with a life-limiting neurodevelopmental disability, the findings of this study are consistent with previous research. Brett (2002) suggests that the extraordinary activities and engagement of these mothers transforms the mothering role experienced by the majority of women with typically developing children. The complexity and nature of the care-work described by mothers in this study is similar to that reported in previous research involving children dependent on technology (O'Brien, 2001; Heaton *et al*, 2005), children with life-limiting and life-threatening conditions (Quinn *et al*, 2005; Steele & Davies, 2006; Manaseri, 2008; Ouelette, 2009), young children with complex life-limiting neurodevelopmental disabilities (Redmond & Richardson, 2003) and children with complex needs (Nicholl, 2008). The consistency of these findings would, again, appear to support a non-categorical approach to exploring the needs of these children, however the unrelenting nature of the work, the complexity of the care required, and the need to be constantly alert and vigilant for changes in the child's condition is not reported in children's disability literature. This sets these children apart from

the group of children with general disabilities, and would suggest that in terms of the labour involved, the care of young children with life-limiting neurodevelopmental disabilities is more closely aligned to that required by children with life-limiting conditions and complex medical needs than to the general groups of children with disabilities. The added dimension of mothers' reports that the complexity and intensity of the child's care requirements makes obtaining outside help difficult is consistent with reports for children with complex healthcare needs (Yantzi *et al*, 2006; Nicholl, 2008, Ouelette, 2009), but again is largely lacking in children's disability literature.

An additional feature of this care-work for young children in this study was the fact that it was continuous 24-hours a day. Mothers reported treatment regimes that often needed to be implemented overnight, and even when no specific overnight interventions were required mothers reported sleep disruption associated with the need for constant vigilance and observation of the child. Sleep disruption is common, and often persistent, amongst children with disabilities (Lancioni *et al*, 1999; Wiggs, 2001; Diddens & Sigafos, 2001; Ryan *et al*, 2002; Coururier *et al*, 2005) and is generally classified as being associated with either behavioural or physical problems. A third type of sleep disturbance, similar to mothers' reports in this study, has been reported in studies of technology-dependent children and is associated with the administration of treatment, management of technology and attention to alarms (Townesley & Robinson, 1999; Heaton *et al*, 2005; Heaton *et al*, 2006). Mothers' descriptions of sleep disturbance, and in some instances chronic sleep deprivation, emphasises the need to address this as a serious problem. This is supported by the fact that sleeping difficulties was the only child-related problem significantly associated with negative family impact score in Stage One of this study. Although chronic fatigue and parental exhaustion is reported in the literature associated with caring for children with life-limiting conditions the impact of this has not been explored. However, the cost and shortage of trained individuals, especially at night, is likely to negatively impact on this important support service for families (Chaguturu & Vallabhaneni, 2005; Heaton *et al*, 2005).

Meltzer and Mindell (2006) report that sleep in caregivers of children with chronic illnesses, particularly children assisted by technology, is significantly disrupted resulting in chronic partial sleep deprivation due to the level of attention and care required by their children. Meltzer, Boroughs and Downes (2010) report that caregivers of technology-dependent

children in their study with limited or no night nursing coverage were not only sleeping about one hour less than caregivers with night nursing, they were averaging six or less hours of sleep per night, the critical limit for daytime performance (Dinges *et al*, 2005). This study also reports an inverse relationship between the total number of nursing hours and the number of urgent care visits for children. The impact of fatigue on areas of general performance and functioning, in areas other than parenting, are well documented where it has been linked to poor concentration, planning and decision-making difficulties (Hockey *et al*, 2000; van der Linden *et al*, 2003a, Torres-Harding & Jason, 2005; Nilsson *et al*, 2005). The lack of appropriate respite described by mothers in this study, especially at night, may have negative effects not only on the mother but also on her ability to care for her child. Aday and Wegener (1988) report that, in terms of daytime functioning, caregivers who received less than 32 hours of nursing care per week reported more feelings of overburden and social isolation. While Van Dongen *et al* (2003) have identified that after 18 hours of cumulative wakefulness, significant performance declines and impairment of executive functioning abilities such as decision-making and flexible thinking occur. It is likely that this adds to the stress experienced by mothers of young children with life-limiting neurodevelopmental disabilities where complex care and complex decision making is an intrinsic feature of the ever-day care provided to their child.

Although this study did not attempt to quantify the work involved in caring for a young child with a life-limiting neurodevelopmental disability, it is obvious from mothers' narratives that this is considerable, consistent, and multi-dimensional. Consistent with previous studies mothers provided intensive, highly skilled, and complex health care to their child as part of everyday nurturance. In order to achieve this they had developed a variety of often complex skills related to the care of their child. These included sophisticated technical and procedural skills, skills of assessment, intervention and evaluation, organisational and planning skills, and advocacy skills. Despite the complex and highly skilled care required by the children in this study, many mothers reported that they did not feel that they were given sufficient time or training to allow them to feel confident and capable with regards to this aspect of their child's care prior to their discharge from hospital. Kelly *et al* (2008) suggest that the transition to optimal home care is more likely to occur if the child's complex needs are organised, the family is trained, adequate management resources are in place, and the

intensity of care coordination needs are reduced. When measure against this standard it is obvious that this was not achieved for the families in this study.

Many studies describe the complexity of the learning curve for parents when a child with complex needs is discharged from hospital (O'Brien, 2001, Alexander *et al*, 2002, 2005; Steel & Davies, 2005; Quinn *et al*, 2005). Harrigan *et al* (2002) proposes that meticulous training for discharge is an essential element of quality of care for all families caring for a medically fragile child. Despite this, studies consistently show that discharge training for parents is often overlooked, and that parents' assumption of the responsibility for highly complex or technological care is not openly negotiated with health professionals (Kirk, 2001; Kirk & Glendinning, 2004; Margolan *et al*, 2004; Nicholl, 2008).

9.4 Families of Young Children with Life-Limiting Neurodevelopmental Disabilities

Reviews conclude that these families caring for children with complex disabilities are physically, emotionally, socially and financially stressed, anxious about their child's condition, and fearful of potential complications and potential emergency situations that may occur both inside and outside of the home (Harrigan *et al*, 2002; Ratcliffe *et al*, 2002; Wang & Barnard, 2004; Nicholl, 2008). In this respect the findings of this study are consistent with previous research exploring the family impact of providing care to children with life-limiting or life-threatening illnesses. Consistent with previous research the findings demonstrate that providing home-care for young children with life-limiting disabilities has impacts far beyond the child and the care they require. The findings emphasise the negative impact that the child's key illness demands can have on the family, and the positive impact that families can have on illness management.

9.4.1 Managing Family Life Day-to-Day

For mothers in this study the most important strategy for managing family life day-to-day was developing and adhering to a strict routine. This was critical to ensuring that the needs of all family members were attended to, and to establish a sense of stability which allowed the family to continue to function effectively. The needs of the child took precedence over all

other aspects of family life and was the fulcrum around which family life pivoted. There was no spontaneity in family life for the families in this study. The care requirements of the child and unpredictable nature of the child's condition necessitated that all elements of day-to-day life needed to be planned in advance so that the necessary resources (medical, technical and personnel) would be available.

This strategy appears to conflict with traditional theories of family normalisation in chronic childhood illnesses. Knafl and Deatrick (1986) laid the foundation for most of the work on normalisation in nursing literature. More recently, following a comprehensive review this was revised to include the following critical concepts: acknowledging the condition and its potential threats to lifestyle; adopting a "normalcy lens" for defining the child and family; engaging in parenting behaviours that are consistent with the normalcy lens; developing a treatment regime that is consistent with the normalcy lens; interacting with others based on a view that the child and family is normal (Knafl & Deatrick, 2002). Deatrick *et al* (1999) note that normalisation is both a cognitive (defining) and behavioural (managing) strategy for parents, with the awareness of the difference between their own situation and the "normal" reference group forming the basis of the concept. Not all of these features were evident in mothers' narratives in this study. Knafl and Deatrick (2002) suggest that families who focus on the normalcy of their situation typically adopt a flexible approach to carrying out the treatment regime, one that emphasises incorporating the illness into the usual routines of the family and child. However the inverse appeared to be the case in this study. Rather than incorporating care into the usual family routines, "normal" family life appeared to be redefined in the context of the child's treatment regimes and needs. It may be that since much of the research on normalisation has been conducted in the context of chronic childhood illness this context does not necessarily fit with the situation of families of young children with life-limiting neurodevelopmental disabilities. Chronic childhood illness may have been more stable over time, and possibly did not involve the type of complex daily treatment regimes that the children in this study required on a day-to-day basis. In addition, the consequences of adapting the child's treatment regime may not have had the same potentially catastrophic consequences for children with more stable chronic conditions. A further point of departure with traditional theories of chronic childhood illness is that mothers in this study did not interact with others based on a view of the child and family as normal. This would have been impossible in the context of mothers' descriptions of how their "difference" was

often reinforced in social situations and interactions with others. This left mothers in the position of acknowledging their family as different from the norm reference group, but accepting that the situation was “normal for us”.

Alternatively, in the context of childhood cancer, Robinson (1993) and Clarke-Steffen (1997) describes a strategy of normalisation whereby mothers construct and live a story of “life as normal”. These researchers describe a process whereby mothers effectively constructed a new concept of normal, characterised by a new routine, so that instead of the treatment regimes being accommodated and manipulated into pre-existing normal family life, normal family life was manipulated to accommodate the treatment regime and care requirements of the child. This concept, in which families individualised and modified the meaning and role of normal activities to fit the particular circumstances and uncertainties of their own lives, better fits the strategies described by mothers in this study. Rehm and Bradley (2005) describe a similar strategy employed by mothers of children who are medically fragile / technology dependent and developmentally delayed. This would appear to suggest that for many families normalisation and feeling or acting normal is not necessarily an all-or-nothing phenomenon, and that the concept is not concrete but may be applied differently in particular family contexts. It also suggest that the process of normalisation for families of young-children with life-limiting neurodevelopmental disabilities more closely resembles that described for families of children with life-threatening conditions and technology-dependence, rather than families of children with chronic conditions or the general group of children with disabilities.

This is supported by the similarities between mothers in this study and those of children with cancer in Clarke-Steffen’s (1997) study. Clarke-Steffen (1997) describes an engagement process which included managing the therapeutic regime, reorganizing roles, evaluating and shifting priorities, and assigning meaning to their situation. All of these features were evident in mothers’ narratives. However, mothers in this study demonstrated an additional feature of the normalisation process whereby they attempted to reinforce their perception of normality through peer support. This involved actively seeking out and engaging with families similar to their own thus bolstering the perception that their family life was not unique and that there were other families like theirs. This strategy has not been reported in previous studies of childhood chronic or life-threatening illness.

9.3.4 Living with Uncertainty

The concepts of uncertainty and unpredictability loomed large over mothers' descriptions of family life in this study. Although it appeared to vary in magnitude, intensity and saliency depending upon the child's condition, living under conditions of sustained uncertainty was part of the tapestry of everyday life for families in this study. It was pervasive and stressful for mothers, one of whom described it as the most difficult element of the care of their child.

Uncertainty has increasingly been identified as an important construct in the clinical and empirical literature on families' responses to serious and chronic childhood illnesses. Mishel's original Uncertainty in Illness Theory [UIT] (Mishel, 1988) indicated how individuals (adults) appraise illness-related stimuli to create meaning in illness. Illness uncertainty is defined as "a cognitive experience elicited in situations in which the meaning of the illness related events is unclear and outcomes are unpredictable" (Mishel, 1990:258) and is comprised of four components including a sense of ambiguity concerning the state of the illness, complexity regarding treatment, lack of information regarding the seriousness of the illness and its prognosis, and perceived unpredictability of the illness course (Mishel, 1984). Uncertainty surrounding the nature (symptoms) and course (prognosis) of a child's chronic condition has been identified as a major stressor that influences family stress, the structure and functioning of the family system, and parental psychological and physical morbidity (Patternson & Garwick, 1994; Rolland, 1994; Cohen, 1993, 1995; Stewart & Mishel, 2000; Dodgson *et al*, 2000; Garwick *et al*, 2002; Santacrose, 2003; Holm *et al*, 2008). It has been reported that the degree of uncertainty associated with unpredictable symptoms in a cohort of young children with chronic life-limiting conditions caused more family distress than the degree of uncertainty of the child's life-expectancy (Dodgson *et al*, 2000).

In her theory of managing sustained uncertainty in childhood chronic life-threatening illness Cohen (1993, 1995) suggests that the diagnosis of a childhood chronic life-threatening condition causes parents assumptive, taken-for-granted, world to abruptly cease to exist. In this context Cohen (1993:83) proposes that parents move from "the secure world of the known, the familiar and the predictable to a normless world of ambiguous boundaries, unclear rules, probabilistic predictions, and sinister possibilities". Cohen (1993) suggests that the management of uncertainty in this situation involves developing strategies to manipulate

the known, the unknown and the unknowable, whereby parents manage six interactive dimensions of daily life: time, social interaction, information, awareness, illness and the environment. Many of these strategies were evident in mothers' descriptions of managing family life in this study. However Cohen's (1993) description of managing the time dimension of sustained uncertainty did not fit with mothers reports in this study. Cohen (1993) suggests that for parents of children with chronic life-limiting childhood illnesses thinking about the future is to fight the threat of loss. Thus she suggests, parents are virtually tied to the present and very proximate future, proposing that parents come to realise that by adopting a one-day-at-a-time philosophy, and living in shortened time units, the perception of uncertainty can be reduced. This particular strategy for managing sustained uncertainty was not apparent in this study, for although mothers were very focused on the day-to-day management of the child's care and needs, they also frequently expressed fears and anxieties related to future issues. These were generally associated with potential service reductions or withdrawals, or worry about what would become of the child in the future. This represented a significant departure from Cohen's (1993) theory. Uncertainty, as experienced by mothers in this study, had an extra dimension. It included not only uncertainty associated with the child's condition itself, but also uncertainty about the future availability and reliability of support services to assist families to meet their child's ongoing care requirements. It is possible that since most of the families in Cohen's (1993) study were caring for a child with cancer the issue of service reduction or withdrawal was an unlikely worry for these families. There was also the possibility that these children would make a full recovery. Families in this study did not enjoy the assurance that the services they required would be available in the longer term, nor was it likely that their need for services would reduce over the course of the child's condition.

Cohen (1995) later investigated the triggers of heightened uncertainty in chronic life-threatening childhood illness and reported seven commonly occurring events which heightened parents' awareness of the uncertainty and anxiety concerning their child's condition and survival. These included medical appointments, bodily variability, keywords and provocative questions, changes in the therapeutic regime, evidence of negative outcomes, new developmental demands and nighttime. It is interesting that none of these triggers were present in mothers' narratives in this study. So while Cohen (1995) proposes that uncertainty ebbs and flows and can be heightened by triggers, mothers in this study demonstrated a

more pervasive and unrelenting form of uncertainty, which like parents in the study by Dodgson *et al* (2000) was associated more with erratic and unpredictable changes in the child's condition than with the child's overall prognosis, augmented with the worry that the services and supports they required to assist them in their caring role may not always be available to them.

9.4.4 Social Consequences for the Family

Disrupted social functioning and increased isolation due to caregiving responsibilities had been reported in many studies of families of children with life-limiting conditions and complex needs (Carnevale *et al*, 2006; Montagnino & Mauricio, 2004; Wang & Barnard, 2004). Rehm and Bradley (2005b) suggest that families of children who are developmentally delayed and medically fragile search for safety and comfort in social situations. They propose that this involves seeking social and physical environments and interactions that assure the physical and emotional safety and comfort of the child and the other family members. Achieving this was very difficult for mothers in this study, with mothers' describing social barriers that could be classified as environmental, child / care related, and attitudinal. As a consequence social opportunities for all family members were severely limited.

Poston *et al* (2003) suggest that social well-being includes the realms of social acceptance, social relationships and social support. Rehm and Bradley (2005b) use the term social consequences to indicate outcomes related to the ability of families to engage in relationships with people outside of close family and to participate in recreational activities. Several factors have been identified as causing negative social consequences for families of children with complex conditions or technology dependence. These include the difficulty of finding competent, skilled respite caregivers and the financial strains that complex chronic or life-limiting conditions impose on the family (Kirk & Glendinning, 2004; O'Brien, 2001; Ratliffe *et al*, 2002; Wang & Barnard, 2004). O'Brien (2001) suggests that the time consuming demands of the child's care and a focus on intra-familial needs results families experiencing social isolation as it impairs social relationships and opportunities for socialisation with friends. Poston *et al* (2003) mentioned the discomfort of friends of family members regarding a child with a disability, while Kirk and Glendinning (2004) cite the heavy and cumbersome equipment that children need, the requirement to return home from activities to start

overnight therapies, and the reactions of others to care procedures as impediments to family socialisation. All of these features were evident in mothers' reports in this study, with mothers' reporting social opportunities limited by the complex care and treatment regimes required by the child, the lack of understanding and sometimes embarrassment they encountered in social interactions with friends, and the perception of the child as a social curiosity when out and about in general.

9.5 Services for Young Children with Life-Limiting Neurodevelopmental Disabilities and their Families

In a series of reviews of children who are medically fragile or technology dependent Harrigan *et al* (2002), Ratcliffe *et al* (2002), and Wang and Barnard (2004) conclude that families need extensive, continuous services including emotional support and skilled supportive and respite care. Families' perceptions of the supports and services provided to them has been identified as a critical factor that influences family quality of life (Feldman & Werner, 2002; Summers *et al*, 2007; Werner *et al*, 2009). These services and supports should both reduce the negative effects produced by the difficulties in the situation (Summers *et al*, 2005) and strengthen the positive effects through interventions favouring family autonomy and empowerment (Soresi *et al*, 2007). Ultimately it is the effects that such services and supports have on children and families that determine the quality and effectiveness of any service or programme.

Current health policy actively promotes primary care for children with life-limiting neurodevelopmental disabilities regardless of the severity of their condition, or the social, economic or cultural circumstances of the family (DOH&C, 2005, 2010). Consequently, at the very least, service delivery should ensure that families are not traumatised by the process of providing care for these children. Consistently mothers in this study referred to the stress and exhaustion caused by the perceived necessity to fight for services, cope with humiliating or disrespectful bureaucracy and regulations, and otherwise deal with their relationships with professionals.

9.5.1 The Efficacy of Current Services

Mothers in this study differentiated between hospital-based services, which were accessed episodically, and the community based services they encountered day-to-day in the care of their child. Particular difficulties were encountered in the hospital context which have been described in Chapter Six, this section discusses the common and general difficulties that families experienced in relation to the services their children received. It is also important to state that mothers reported that they were “fairly satisfied” with the individual services that they received, and the personnel they encountered in these services, when they finally managed to access a service. Accessing services was not a binary outcome. It was not a question of children receiving services or not. Instead there appeared to be a variety of potential challenges that parents encountered associated with the process of obtaining and keeping the services that are needed.

The principal difficulty encountered by mothers was in accessing services in the first place. Mothers’ reported that this was due to a combination of factors including lack of essential services, inconsistent service provision, inequitable service provision and resource allocation, and lack of information about services. Service providers agreed that these difficulties are commonly encountered by this population of children and their families. In addition to the problems of insufficient, under-funded and under-resourced services, mothers’ also identified several areas of process deficiencies and inefficiencies relative to the services they received. These included service integration and coordination, continuity, and relationships with health professionals.

9.5.2 Service Integration and Coordination

The children in this study received care from a number of different services and agencies. It would appear that when families receive care from a variety of sources, connecting that care into a smooth trajectory becomes increasingly difficult. Mothers’ described a tendency for programmes, agencies, even disciplines to function in isolation. The consequences of this were that they experienced service fragmentation and inefficiencies and a perceived failure to see the needs of the child in a holistic way, although services providers did not concur with this opinion.

The World Health Organisation (2008) identifies five common failings of health care systems which policy makers should be aiming to address. Amongst these are the problems of fragmented and fragmenting care with no coordinated approach and a lack of continuity of care. Almost two decades before this the Department of Health and Children (2001b) identified several weaknesses in the Irish health system including inadequate linkages between services and fragmentation of services. Although Irish health policy reports consistently urge a concerted effort to avoid fragmentation and enhance continuity of care (DOH&C 2001a, 2001b; DOH&C, 2004b) it would appear that such efforts have largely been ineffective.

It may be that efforts to formulate solutions have been hampered by a lack of consensus in the literature about the meaning of the terms “service coordination” and “service integration” with poorly defined terms that are often used interchangeably. Park and Turnbull (2003) suggest that service coordination has been viewed as a systematic process for assisting families in obtaining the services and resources they need, while service integration, a more recently introduced term, is essentially an extension of this concept. They propose that service integration relates to a systematic effort to provide appropriate and harmonized service to young children and their families based on collaborative partnerships between families and professionals, among professionals and among agencies that are formed in the process of enhancing child and family outcomes. Alternatively King and Meyer (2006) proposed that service coordination and integration are distinct but related concepts. These authors suggest that service coordination is essentially a clinical function that brings different services into a efficient relationship for a given family, while service integration deals with the organisational perspective and relates to functions and activities aimed at the formation of a comprehensive range of services in a geographical area where the intent is to enhance the effectiveness of service delivery. It would appear that both were problematic in this study. Regardless of the different conceptual analysis there is agreement that the goals of both are to enhance the likelihood that families perceive care to be easy to access, seamless and tailored to their needs (Park & Turnbull, 2003; King & Meyer, 2006). While integration does not automatically improve services, it can be a facilitator of improved quality if it is used to deliver services in a more ‘useful’ way (DOH&C, 2004b).

Sia *et al* (2004) advise that delays, gaps, duplication and diffused responsibilities which characterize fragmented care are expensive, inefficient and sometimes hazardous to health. Coordinated care has been proposed as a solution to this problem (Alexander *et al*, 2004; Hefener, 2010). Coordinated care is defined as linking patients and families to services and resources across many subsystems of the health and human service fields (Gupta *et al*, 2004). However Alexander *et al* (2005) identify several barriers to coordinated care encountered by children with complex health care needs including: a lack of knowledge and information about community resources; a lack of communication among health care professionals and organisations involved in the child's care; and a lack of clearly defined roles for each of the agencies involved in the child's care. Many of these features were evident in mothers and service provider's accounts in this study.

The majority of mothers reported experiences of fragmented services, with several factors identified as contributing to this experience. The most obvious of these related to access and information difficulties. This appeared to be most marked in the context of community based services where mothers reported considerable difficulties associated with accessing both information about services and indeed services themselves. A second major contributing factor to mothers' experiences of fragmented care related to the acute care / primary care interface. This has been widely reported in previous studies exploring parents' experiences of caring for children with life-limiting conditions or complex needs (Margolan *et al*, 2004; Kirk & Glendinning, 2004; Quinn *et al*, 2005). In this study this resulted in mothers being discharged from hospital unprepared and unsupported, and often without the support of available community based services. It is likely, given the opinion of services providers, that hospital personnel's lack of knowledge of locally available community based services, and poor communication between acute and community services, contributed to this fragmentation.

9.5.3 Continuity of Care

Parents in this study also reported difficulties associated with continuity of care for their children. Although Reid *et al* (2002) suggest that continuity of care means different things to different caregivers, and definitions are often presumed rather than stated, there appeared to be two principal components of continuity that were considered deficient by parents in this

study. The first of these related to continuity of staff and relationships. Relational continuity was explicitly stated as a factor that did not work well in current services by parents in stage one's survey. Parents' follow-up interviews suggested that relational continuity was important for several reasons: in facilitating a trusting relationship; allowing the professional to get to "know" the child, which mothers considered important for the provision of holistic care; and in facilitating a more equal partnership between the mother and the professional.

Continuity of relationships has been found to be important in previous studies of parents of children with life-limiting conditions (Kirk & Glendinning, 2004; Heller & Solomon, 2005) where it was equated to increased confidence about the quality of care delivered. Kirk and Glendinning (2004) suggest that continuity in parent-professional relationships facilitated relationships characterised by mutual recognition of respective knowledge and expertise. Heller and Solomon (2005) concur, concluding that continuity of care to this population of children and their families encourages sharing of expertise and information. Christakis *et al* (2002) suggest that continuity of care is associated with service satisfaction in primary care services for children. In addition to shared mutual knowledge where there was continuity of relationships parents reported that they were more likely to feel respected and listened to (Christakis *et al*, 2002).

The second issue related to continuity that mothers' described was associated with continuity of information. Although not specifically identified in the stage one's survey (parents did however report communication difficulties), this was a consistent feature of mothers' narratives, and was a cause of considerable frustration for mothers. It would also appear that when service providers agreed that there was poor communication between acute and community based services that poor continuity of information was the principal issue referred to in this context. Reid *et al* (2002) suggest that the ways providers use and transfer information is critical to adapting care to meet the needs of the individual, as it bridges separate elements of care over time and is a prerequisite for coordinated care. Informational continuity was problematic at all levels for mothers in this study. It occurred between different disciplines in the same services, between acute and community based services, and between different services working the community.

9.5.4 Parent-Professional Relationships

Dealing with health services and health professional was an integral part of providing care to their children; however mothers in this study did not only report difficulties associated with the integration and coordination of services. They also wanted service providers to listen to them and respect what they had to say, treat them with courtesy, and respect their expertise in the care of their child. Parents of children who have disabling conditions often define their relationships to include the professionals who work with their children (Chomicki *et al*, 1995; Seligman & Darling, 2007), subsequently professionals need to recognise the importance of interacting with families beyond the provision of direct services (Smith Stepanek, 2008).

The way in which professionals support children and their families has the potential to enhance or impede family outcomes (Dempsey & Keen, 2008) yet despite the family-centred care philosophy that has been adopted, at least rhetorically, by most children's services, relationships with professionals were often difficult for mothers in this study. Mothers' described what they perceived as a lack of emotional support, lack of empathy, and an apparent failure to respect, or even acknowledge, their expertise and experience. Mothers' perceived ambivalent position was supported by the opinion of the expert panel in relation to the service providers' perceptions of parents of young children with life-limiting neurodevelopmental disabilities.

For several decades family-professional partnership has been recommended as a critical feature of the delivery of effective services to children. A commitment to partnership practice underpins all Irish health and child care policy with several policy documents produced by the Department of Health and Children advocating a commitment to the principle of effective partnership with parents (DOH&C, 1999, 1999a; 2000; 2002; 2004b). The DOH&C (2004b:15) proposes a bi-dimensional concept of partnership to include the relationship between the service and families, and the relationship between agencies and disciplines engaged with a role in helping to meet the needs of the family. With regard to the former, they state that "effective partnership practice involves a commitment to the provision of information, practical arrangements and emotional support to parents engaged with services". The quality of parents' partnerships with service providers is a critical element of their overall quality of life (Blue-Banning *et al*, 2000), and is a crucial factor in the delivery of services to achieve the best possible outcomes for children and their families (Park &

Turnbull, 2003; McIntosh & Runciman, 2007). Research indicates that parents and professionals alike define collaborative partnerships at least in part in terms of the quality of their interpersonal relationships with each other (Summers *et al*, 2001; Park & Turnbull, 2003). Based on data from 33 focus group interviews and 32 individual interviews with parents and service providers Blue-Banning *et al* (2004) propose six inter-related themes of collaborative family-professional partnership in the context of childhood disability including communication, commitment, equality, skills, trust and respect. Trust and respect were also identified as core features of family-professional partnerships by parents of children with disabilities in Fereday *et al*'s study (2010). Parents highlighted that they had a different, but equally valid, knowledge of the child, and when this was respected a trusting relationship developed.

Kirk (2001) suggests that parental expertise in the care of a child requiring complex care can transform the nature of the parent-professional relationship as well as the roles of the professionals themselves. Previous research suggests that parents are often dissatisfied because they do not feel that they are respected as partners who have parental expertise and competence (Kirk, 2001; Balling & McCubbin, 2001; Fisher, 2001; Kirk & Glendinning, 2004). While Taylor (2000) suggests that in hospitals the power ratio favours hospital staff with parents disadvantaged as visitors in an unfamiliar environment, Hewitt-Taylor (2005) proposes that this situation is reversed in the context of home-care. However changes in the balance of power do not necessarily lead to the development of parent-professional relationships that are characterised by partnership, and interactions with health professionals and service providers can act to either increase or reduce parents coping (Hauskov-Grungaard *et al*, 2011).

It may be that that health-professionals are unsure of the boundaries of the family-professional partnership. Blue-Banning *et al* (2004) reported that although the themes of partnership were agreed between parents and professionals one area of difference was in the emphasis placed on commitment and equality. While parents wanted professionals who would go the extra mile, professionals expresses reservations about taking these concepts too far. These reservations centered on professionals perceived need to empower families and concerns about fostering co-dependency and actually causing harm to the family. Regardless of the reason genuine parent-professional partnership was not evident in this study, and

mothers perceived this as an additional source of stress. In the hospital environment mothers described being left to provide most of the hands-on care and observation that their child needed, while in the home environment mothers described being left to get on with providing care often with little support. This was not empowering for these mothers, on the contrary it acted to increase their stress and burden. In addition mothers were rarely invited to participate in the planning of care for their child, nor were they involved in any meaningful way in the evaluation of the services provided.

9.6 Expressed Unmet Service Needs of Young Children with Life-Limiting Neurodevelopmental Disabilities and their Families

Two decades have passed since Woolley *et al* (1991:216) interviewed parents of children with chronic life-threatening conditions and first described what parents experienced as “the complex and often distressing job of obtaining help”. Almost every study that has been conducted in the intervening years has highlighted the need to develop significant instrumental supports for these families, and the findings of this study are no different. Mothers identified several unmet support needs in this study. These included the need for information, better respite, and a need for a key-worker to organise and coordinate care.

9.6.1 Informational Needs

The complex and difficult task of accessing information arose as a consistent finding in this study. Providing general information received the lowest mean score on the MPOC subscales in stage one, where a single source of information was also identified as a factor considered necessary to improve services. Mothers’ narratives described the difficulty experienced in obtaining information, and the reliance on others parents as a source of vital information about services and entitlements. Service providers unanimously agree that accessing information is a difficulty for parents.

Information is critical to families of children with chronic or complex conditions as a resource in the management of the child’s condition, a means of adjusting to the future, as a method of establishing control, and as coping resource (Cohen, 1993; Hummelinck & Pollock, 2006; Nuutila & Salanterä, 2006; Smith Stepanek, 2008; Kanpp *et al*, 2010b).

Despite this, previous research suggests that accessing information is a consistent challenge for parents of children with life-limiting and life-threatening conditions, with access to information arising as a consistent theme in research literature related to parents' needs (Lenton *et al*, 2001; Kirk & Glendinning, 2002; Redmond & Richardson, 2003; Quinn *et al*, 2005; Monterosso *et al*, 2007). Most studies describe problems with parents reporting having to seek out information for themselves or receiving insufficient or conflicting advices which leaves them confused.

Research suggests that parents of children with life-limiting and life-threatening conditions require a variety of different types of information. The parents of children with a wide variety of non-malignant life-limiting conditions in Lenton *et al*'s study (2001) reported that they needed more information and specific advice on what to do at times of illness. Similar findings were reported by Monterosso *et al* (2007) by parents of children with cancer. Although mothers in this study reported that they found it difficult to get information on their child's condition at the outset, medical or condition related information was not the greatest informational difficulty they currently encountered, supporting the opinion that the information needs of parents develop and change over time (Hummelick & Pollock, 2006; Nuutila & Salanterä, 2006). Consistent with the findings of Redmond and Richardson (2003) and Quinn *et al* (2005) mothers in this study reported accessing information about support services as one of the greatest on-going challenges they encountered. This included accessing information about therapeutic services, support services and entitlements available to the child and family. Many mothers reported having to depend upon peer support to provide this information, and in some instances the lack of formal information meant that families were not availing of services or supports to which they were entitled. This difficulty was acknowledged by the expert panel of service providers. The agreement of service providers that acute services are often unaware of the services available to young children and their families in the community suggests that service providers themselves contribute to this difficulty.

9.6.2 Need for Additional Support Services

In addition to difficulties related to the process of service delivery, mothers also identified specific support services which were needed but which were either unavailable or inflexible

to the needs of the family. In many cases in-home respite and support services, while very gratefully received, were not necessarily sensitive to the family's needs. Mothers' reported that they had to avail of respite (either in or out of home) when it was available rather than when it was most needed or most beneficial to the family. In addition they also reported confusion about the role of in-home support workers.

Respite care is a vital element in the continuum of care for children with chronic life-limiting conditions, especially for children with medium to long-term high dependency needs (Quinn *et al*, 2005; Fowler-Kerry, 2008; DOH&C, 2010) where it can provide families with a temporary reprieve from the burden of care and an opportunity to spend time together as a family away from the stress of everyday caring (Horsburgh *et al*, 2002). In addition, an accessible respite service can reduce the need for hospital admission by providing the community supports necessary to sustain parents in their caregiving role. In a review of repeated admissions of children with chronic health problems Kelly and Hewson (2000) report that 20% of hospital admissions were due to the lack of appropriate community support services including respite for parents. The Canadian Association for Community Care (2002:5) describes the provision of adequate respite care as an "absolutely essential aspect of home care" and calls for a complex system of respite services to meet the wide and diverse needs of families of children with complex needs who they describe as "heavily burdened". Similarly the European Association of Palliative Care [EAPC] standards for the development of children's palliative care suggest that respite for family carers and the child is essential and should be provided in a flexible manner (Craig *et al*, 2008). In the context of severe intellectually disability several factors influence the use of respite care (Chan & Sigafos, 2000), with exhaustion, both physical and mental, being one of the principal reasons for a carers use of respite services (Hoare *et al*, 1998; Hartrey & Wells, 2003; Eaton, 2008). Parents who express a need for respite are usually severely stressed and caring for severely disabled children, although this does not appear to influence the frequency or ease of access to respite provision to these families (Neufeld *et al*, 2001; Chadwick *et al*, 2002; MacDonald *et al*, 2006; Doig *et al*, 2009).

There is little literature evaluating the impact of respite on family caregivers of children with life-limiting conditions. In the context of children with disabilities reduction in stress levels

and increased psychological well-being are reported (Cowen & Reed, 2002; Shu *et al*, 2002) as well as positive effects on family functioning (Chan & Sigafoos, 2001; MacDonald & Callery, 2004). In a general review of the research literature Chan and Sigafoos (2001) suggest that, at least in the short-term, respite care is associated with significant reductions in parental stress for the majority of parents who use it. Alternatively the adult literature suggests that when respite is offered too late or used too little in the caregiving experience caregiver burden and negative health outcomes are not reduced (Zarit *et al*, 1999, Zarit, 2001).

MacDonald and Callery (2008) describe a trajectory of care for intellectually disabled children requiring complex care which suggests that respite needs increase over time as the child gets older. In this study, despite the young age of the children in this sample, mothers expressed a current and urgent need for greater respite services, a situation consistently identified in previous Irish studies of children with life-limiting conditions and disabilities (Redmond & Richardson, 2003; Quinn *et al*, 2005). Despite this expressed need the issue of respite was complex for mothers. Only two families availed of out-of-home respite services (a third was on a waiting list) and when mothers discussed the need for better respite facilities this generally referred to in-home respite. Respite was highly valued by those families to whom it was available, and was identified by those to whom it was not as a service that was required to make family life easier. Many mothers described a general lack of available and appropriate respite, which was compounded by the complexity of the child's care requirements which made obtaining appropriately qualified carers difficult.

Although respite is generally considered to be a relief from the burden of care provision, Kerry-Fowler (2008) offers an additional perspective on its role. Mothers of children with life-limiting and life-threatening conditions in this qualitative study described how, in addition to providing much needed relief, respite allowed them an opportunity to actualise themselves more in the role of parent rather than the role of primary caregiver. A similar perspective was described by mothers in this study. This was especially true where there were other children in the family as in this context respite allowed the mother to spend some relatively uninterrupted and quality time with the child's siblings. Subsequently the provision of sufficient and appropriate respite has the potential to impact positively not only on the

mother as the primary caregiver, but also on the siblings of the young child with a life-limiting neurodevelopmental disability.

Most reviews in the area of respite care find flexible definitions of respite and a range of services included (Olsen & Maslin-Prothero, 2001; Cheeson & Westwood, 2004; Merriman & Canavan, 2007). The definition and purpose of respite also seems to depend upon who is asked (Olsen & Maslin-Prothero, 2001; MacDonald & Callery, 2004). Zarit (2001) proposes a broad interpretation of respite services suggesting that respite encompasses all programs and services that provide temporary relief to family caregivers. Within this interpretation all in-home support services (including home support workers and home-helps) effectively provide a form of respite care. It is difficult to compare general in-home support services with published literature in the context that the types and availability of such services vary greatly and beyond being acknowledged as a supportive service to parents there is no empirical literature exploring this aspect of services. Several areas of difficulty were identified by mothers in this study in relation to these ancillary support services which included access difficulties, unexplained or sudden service withdrawal or reduction, and role confusion.

Non-professional in-home support was complicated by the fact that often the same individual performed more than one role (home-support worker on one day and home help on another) which caused confusion about particular roles and responsibilities and which tasks could or could not be performed by in-home support workers. In addition mothers described how this service could be reduced or withdrawn without reasonable explanation (beyond vague reference to funding issues) even though their home and family circumstances had not changed.

Respite care can be structured in a variety of ways and be provided both formally and informally, each of which has unique advantages and disadvantages (Merriman & Canavan, 2007). Formal in-home respite was the preferred form for mothers in this study. The same preference was expressed by Irish carers in a number of small-scale qualitative studies (Hartrey & Wells, 2003; Redmond & Richardson, 2003). One of the difficulties with in-home respite is the fact that children with complex conditions requiring specialised care need to

have respite that is provided by a qualified nurse who can respond to the child's changing needs and be competent with the use of any equipment required (Olsen & Maslin-Prothero, 2001; Valkenier, 2002). Currently respite care to these families is provided through a provider based model, with professional carers were provided through either the HSE or through a charity service. This meant that families had to avail of professional respite whenever a suitable carer was available. It may be that a more flexible consumer directed model of respite provision is required to meet the needs of families caring for a young child with a life-limiting neurodevelopmental disability.

Consumer-directed modes of financing and delivering services permit service recipients, as opposed to medical or social work professionals, comparatively greater choice and control over all aspects of service provision (Doty *et al*, 1996; Kodner, 2003). Although various approaches with differing degrees of professional monitoring are possible (Tilly & Wiener, 2001) effectively consumer-directed models of service provision are based upon beneficiaries receiving fiscal resources enabling them to purchase the services they want (Tilly *et al*, 2000). Although this model is not used in any context in Ireland, it is widely used in adult services in several European countries (Tilly *et al*, 2000) and in disability services in America (Heller *et al*, 1999).

The move from the current agency-based model to a consumer-based model of respite provision would move control over who provides the service, and how and when this service is delivered, to the family. The model could also include the ancillary services that form the total respite package that the family receives. In this context families are allowed to hire the support attendant, define the attendant's duties, and decide when and how specific tasks or services are performed (Doty *et al*, 1996) which would eliminate the issues of unsuitable hours and role confusion described by mothers in this study. Although families tend to hire friends, neighbours, and members of their extended families in non-professional support roles (Caldwell & Heller, 2003) this could in fact provide additional benefits for families in that the advantages of family support are retained and the difficulty of asking for help reduced. Evaluation of consumer directed models of service provision indicate that families with more control over services in this way had fewer unmet needs and used more services than the control group; were more satisfied with the services they received; experienced greater self-efficacy, and were less likely to desire an out-of-home placement than the control group

(Heller *et al*, 1999; Caldwell & Heller, 2003). Similar findings were reported by Benjamin *et al* (2000) who report that families in the consumer-directed model report more positive outcomes than those in the agency model, in relation to unmet needs, and service satisfaction. A family member present as a paid provider was also associated with more positive reported outcomes within the consumer-directed model.

Although health professionals have expressed concerns about the capacity of consumer direction to assure quality, particularly with respect to safety, meeting unmet needs, and technical quality, evaluations of the model suggest that these are unfounded and that the consumer-directed service model is a viable alternative to the agency model, producing quality of care that is at least comparable to that provided by the agency-directed model (Benjamin *et al*, 2000; Benjamin, 2001; Tilly & Wiener, 2001).

9.4 Need for a Key-Worker and Improved Multi-Agency Working

Both mothers and service providers indicated a need for a key-worker for all families caring for a child with a life-limiting neurodevelopmental disability. There was however a slight difference between parents and service providers in respect of what functions a key-worker should perform. While service providers focused on the service-coordination function of a key-worker, parents envisioned a much broader role particularly in relation to a key-worker who would also act as a single source of information about services and entitlements. In this respect key-workers represented the single point of contact and information that families would like.

Greco *et al* (2005:v) describe a key-worker as “a named person whom the family can approach for advice about, and practical help with, any problem related to the disabled child” suggesting that the term is often used interchangeably with care coordinator, link-worker, or service coordinator. The value of having one named person who acts to coordinate the input to the family from the various agencies and services involved in the child’s care is well recognised (Cavet, 2007). In a review of the evidence related to key workers Liabo *et al* (2001) concluded that findings indicate that a key-worker service is associated with: improved overall quality of life for families; better relationships with services; better access to services and benefits; and reduced levels of stress. Similar findings about the value of a

key-worker were reported by Townsley *et al* (2004) and Greco *et al* (2005). Additionally good personal relationships between key workers and parents are reported by parents' as important factors and of value in themselves (Beresford, 1995). Sloper *et al* (2006) and Beecham *et al* (2007) report that the extent to which key-workers carry out various aspects of their function and the number of role aspects they perform is a strong predictor of family outcomes.

The topic of key workers for families of children with life-limiting conditions has received considerable emphasis in health policy formulations (DOH&C, 2001; 2005; DOH&C, 2010). In its policy document *Palliative Care for Children with Life-Limiting Conditions* the DOH&C (2010) identify a key worker, specifically a Children's Outreach Nurse, as fulfilling the role of improving the coordination and integration of services and providing continuity of care. Although this is generally considered to be an integral part of a key-workers role (Cavet, 2007) previous research however suggests that it may be an aspirational and overly optimistic aim.

Notwithstanding the evidence in favour of key workers, in practice the simplicity of the idea is contrasted by the complexity of its implementation. In respect of the proposed service coordination function of the key-workers role research suggests that families with a key-worker do not necessarily experience fewer problems with services than families without a key-worker (Beresford, 1995, Greco & Sloper, 2004). It would appear that, while effective in an informational and supportive role, the effectiveness of a key-worker in reducing service related problems is ultimately dependent on interagency cooperation and service availability (it is not possible to coordinate services that do not exist in the first place). Greco *et al* (2005) suggest that if key worker services are to be part an effective service system, implementation must take place on an inter-agency basis. The efficacy of the key worker's is therefore dependent on a basis of good multi-agency working at both strategic and practice levels. This was not evident in this study with mothers reporting poor coordination and integration of services, and poor communication across the acute / primary care interface, and both within and between agencies. In the context of children with complex healthcare needs, Watson *et al* (2002b) suggest that there is a continuum of joint working ranging from multidisciplinary to transdisciplinary working. They suggest that transdisciplinary working is "a synthesis of services" (p.53) whereby packages of care and support are developed to meet the particular

needs and aspirations of individual children and their families. This level of working was not evident in this study, for although there was evidence of multidisciplinary working, mothers' reported little coordination between agencies and service delivery focused on the child's needs which were narrowly defined.

Research on multi-agency working also provides consistent findings on factors that can facilitate or act as barriers to coordination of services (Watson *et al*, 2002b; Cameron & Lart, 2003; Carter *et al*, 2007)). These studies suggest that successful multi-agency working is promoted by: clear and realistic aims and objectives which are understood and accepted by all agencies, leading to a clearly defined model of how the multi-agency service will operate; agreement about how resources will be pooled or shared; clearly defined roles and responsibilities, so everyone knows what is expected of them and of others; clear lines of responsibility and accountability; and ensuring good systems of communication and information sharing at all levels. Factors that hinder joint working include: constant reorganisation; frequent staff turnover; lack of qualified staff; financial uncertainty, difficulties sustaining initiatives when funding ceased and difficulties in ensuring equity from partner agencies; and different professional ideologies and agency cultures (Atkinson *et al*, 2002; Koppel *et al*, 2001; Greco *et al*, 2005). It may be that many of these issues need to be addressed in current services before it can be expected that the introduction of a key-worker for families can improve the coordination and delivery of these services.

9.5 The Intersection of Policy, Theory and Practice

Because of the nature of their conditions children with complex life-limiting neurodevelopmental disabilities occupy a space at the intersection of medical and social policy. The past two decades have seen the models that inform policy for disabled children and their families undergo radical changes based upon the social model of disability which was critical of the traditional individual medical approach in favour of a more socio-political approach (Barnes, 2005). At the same time advances in medical knowledge and care have increased the number, and extended the longevity, of children with complex and life-limiting neurodevelopmental disabilities, a trend that is likely to continue into the future (Goldman, 2003).

Although current policy discussions acknowledge that children with developmental disabilities are a heterogeneous group, policy prescriptions and legislation are aimed at the group as a whole, and focused at the level of the individual rather than the level of the family (Government of Ireland, 1999; 2004; 2004b; 2004c; 2005). While this may be effective at a general level it does not appear to account for the exceptional needs of these children and their families, nor does its individual-level focus accommodate the exceptional demands made on the families of these children. The children with life-limiting neurodevelopmental disability in this study represented a unique and vulnerable subpopulation of disabled children, who have complex medical needs, a requirement for ongoing high intensity care, and utilisation of a wide variety of health services. In this context they differ in significant ways from the broader general group of children with disabilities supporting Hefner's (2010) assertion that these children should be the focus of independent studies and policy discussions in the context of general disability. In addition, while family disability literature over the past decade is critical of a burden-orientated focus on families of children with disabilities (Scorgie & Sobsey, 2000; Greene, 2007a; Trute *et al*, 2010) the findings of this study suggest that not only do families of young children with life-limiting neurodevelopmental disabilities carry a heavy burden and face many challenges and difficulties over the course of their caregiving, but that this burden negatively impact on all dimensions of family life, and on all members of the family unit.

Current health policy prescriptions advance the primary care model for children with complex and life-limiting conditions (DOH&C, 2010), and this is consistent with the wishes of mothers in this study and in previous studies (Redmond & Richardson, 2003). However this appears to be at a significant cost to the wellbeing of primary caregivers and families, and clearly policy does not target families with high service needs in ways that adequately support them in caring for children with life-limiting neurodevelopmental disabilities. In the absence of explicit national best practice guidelines for children's palliative care the DOH&C (2010) adopt the United Kingdom based ACT Charter (ACT, 2003). This charter outlines the basic requirements of children and families receiving palliative care, and includes 14 items related to the delivery of services that such children and their families should expect to receive. These include that every child with a life-limiting condition and their family should expect: to be included in the process of care planning; to be provided with timely and appropriate information; to have access to a key-worker; and to be offered regular and

reliable respite care. It is obvious from the findings of this study that services to young children with life-limiting neurodevelopmental disabilities and families are not in accordance with these best practice guidelines.

There is also the issue of how best to approach research with these children and their families. On one hand the findings of this study support a non-categorical approach to exploring the needs of children with life-limiting conditions. The symptom profile of the children in this sample was comparable with that of children with a wide variety of life-limiting and life-threatening conditions in a study by Lenton *et al* (2001), and the difficulties families described in relation to engagement with services are generally consistent with those in other published literature for more generic populations of children with life-limiting conditions (Hunt, Elston & Gallaoway, 2003; Redmond & Richardson, 2003; Quinn *et al*, 2005). However the findings also suggest limitations to such a non-categorical approach. The complexity and continuous nature of their conditions, and the high level of skill their care demands, differentiates young children with life-limiting neurodevelopmental disabilities from both the general population of children with chronic conditions, and the general population of children with developmental disabilities. In addition research suggests that this population of children may be less supported by extended family than those with less complex needs (MacDonald & Callery, 2008; Monterosso *et al*, 2007). Families in this study crossed the divide between theories of chronic illness, life-limiting illness, and theories of disability, suggesting that this is a unique group within the population of children with life-limiting conditions and that the application of traditional individual models may not encompass this uniqueness. This has recently been recognised within the overall realm of the field of Disability where, within the last few years, scholars within the Disability Rights Movement have begun to critique the social model of disability, debating its generalisability and proposing potential alternatives to the social model (Hughes & Paterson, 1997; Chappell, 1998; Chappell & Lawthom, 2001; Shakespeare & Watson, 2001; Gabel & Peters, 2004; Burchardt, 2004). Chappell (1998) and Chappell and Lawthom (2001) argue that one of the difficulties of the social model of disability is that it attempts to encompass the experiences of all disabled people, and in doing so challenges the separation of disabled people from each other. Similarly Hughes and Patterson (1997) have indicated a concern that the definitions presented by the social model deny that bodily impairment has any relevance, and argue that the realignment of the disability / impairment distinction is vital. Shakespeare and

Watson (2001) also propose that “the "strong" social model itself has become a problem” (p. 13) arguing that a modernist theory of disability which seeks to provide an overarching meta-analysis covering all dimensions of every disabled person's experience is neither a useful nor attainable concept.

9.6 Confirmation of the Study's Theoretical Framework

As described in Chapter One, this study was underpinned by Family Stress, Adaptation and Adjustment Theory and its associated Double ABCX Model of Family Adaptation (McCubbin & Patterson 1982, 1983a, 1983b; McCubbin & McCubbin, 1991, 1993; McCubbin *et al*, 1996a). According to this theory family stress and adaptation to adversity is influenced by the cumulative pre-crisis and post-crisis stressors that the family experience, which interact with the availability of existing and new resources and the meaning that a family gives to a situation, to support the family in responding and coping with adversity. Although the study did not test the individual components of the theory, at a general level there is strong support for the theoretical framework in the findings of this study.

Mothers' descriptions of the birth of their child and initial hospitalisation support the proposition that this has a devastating effect and constitutes a major family crisis. Two specific types of stress were identified by parents in this study. On one hand there was the necessary, or unavoidable, stress of coping with the child's condition and complex care requirements, and the ensuing cognitive, physical and emotional challenges with which the family was confronted. Although inevitable, this type of stress was mentally and physically exhausting for parents due to the nature of the care provided and the unpredictable crises in the child's condition with which they were confronted. This stress was not stable, for although there was the chronic stress associated with the child's care, parents also reported episodes of acute stress associated with changes in the child's condition and episodes of hospitalisation.

On the other hand, parents also reported avoidable or unnecessary stress. This was principally associated with the frequent conflicts and battles they encountered with professionals and service systems. The battle with service systems most often related to an unmet need for care and support services. This has also been reported in previous research (Redmond &

Richardson, 2003; Hunt *et al*, 2003; Quinn *et al*, 2005; Nicholl, 2008). Battles with health professionals were most frequently associated with frustration and a perceived lack of partnership. These findings are generally consistent with previous research exploring the experiences and needs of parents of children who have complex disabilities (Redmond & Richardson, 2003), complex medical needs (O' Brien, 2001, Alexander, 2002; Carnevale *et al*, 2006; Steele and Davies, 2006; Smith Stepanek, 2008), and parents of children diagnosed with rare conditions (Dellve *et al*, 2006). This group of researchers reported that families of children with complex, rare, or poorly understood conditions often do not feel validated by service providers and the unremitting physical and emotional demands that parents experience are often not fully understood by the professionals who play a role in providing for the care needs of these children.

The physical, emotional, social and financial stress reported by parents in this study is consistent with previous research findings (Harrigan, 2002; Ratliffe, 2002; Redmond & Richardson, 2003; Wang & Bernard, 2004, Quin *et al*, 2005; Steele & Davies, 2006). The cumulative stresses experienced by parents in this study had a negative impact on all dimensions of family life, and affected every member of the family unit either directly or indirectly.

Mothers in this study identified several adaptive resources which mediated the experience of stress associated with caring for their child. These could be classified as intra-familial and extra-familial. Intra-familial resources included family relationships and the support of partners, and extended family. In addition mothers' described personal resources such as personal strength, pragmatism and optimism which assisted in their daily coping. This is consistent with the assumption of Family Stress, Adjustment and Adaptation Theory that all families possess strengths and develop competencies to protect and assist in family recovery from both expected and unexpected non-normative stressors following a family crisis. It supports the move to a family strengths and resiliency perspective which has become an important concept in child development and family nursing theory (Patterson, 2002; Blundo, 2001; Allison *et al*, 2003; Walsh, 2006; Bernard, 2006; Sittner *et al*, 2007; Dunst & Trivette, 2009). It also suggests individual resilience on the part of the mother which Patterson (2002) suggests is an important facet of family resilience in the context of theories of family stress. Extra-familial resources were also reported by mothers in this study. The findings support the

premise that social support (in this study principally from other mothers in similar situations) is an important protective factor and adaptive resource (McCubbin & McCubbin, 1991, 1993; McCubbin *et al*, 1996a), and is also consistent with previous research (Dyson, 1997; Black & Lobo, 2008).

Family Stress, Adaptation and Adjustment Theory proposed that services available to children and their families form part of the family's expanded resources, and consequently should act as a protective factor against family stress (McCubbin & McCubbin, 1991, 1993; McCubbin *et al*, 1996a). Unfortunately this was not evident in the findings of this study. Although all parents reported individual professionals who were compassionate, encouraging, and supportive during their stressful journey, in the main parents interactions with service providers in this study acted as a stressor rather than a protective factor. This finding is consistent with previous research reporting parents experiences of caring for their child with a life-limiting illness (Redmond & Richardson, 2003; Hunt, Elon & Galloway, 2003; Kirk & Glendinning, 2004; Smith Stepanek, 2008; Nicholl, 2008). However, mothers' in this study did describe a process of "meaning-making", and consistent with the assumptions of Family Stress, Adjustment and Adaptation Theory this appeared to act as a recovery factor.

As stated in Chapter One this study was not designed to be a test of Family Stress, Adjustment and Adaptation Theory. Consequently critical components of the theory, for example family types and family outcomes, were not measured or explored in the study. The theory did however provide a guiding framework for the study, and the findings suggest that this is a constructive and useful framework for understanding the needs of this population of children and their families, and for framing interventions with families.

9.7 Overall Strengths and Limitations of the Study

This study is unique in its investigation of the palliative care needs of this specific group of children and their families in an Irish context. Despite the attendant difficulties relative to the general complexity of terminology, the study provides the first comprehensive overview of the specific needs and experiences of a well defined group of life-limited children and their families. The attempt to investigate not only the quantification of physical problems

experienced by these children, but the efficacy of their management is a unique feature of the study.

Previous quantitative studies have used various standardised instruments to measure the impact on the family of caring for a child with a life-limiting condition and complex disabilities; however these have missed the rich texture of an experience that is essentially personally lived (Lenton *et al*, 2001; Roberts & Lawton, 2001; Curran *et al*, 2001). In an attempt to rigorously measure specific impacts and aspects of the experience, studies using purely quantitative methods have divided and compartmentalised the experience of caring for these children, and consequently lost the actual context in which this care is delivered and experienced. Conversely, while qualitative studies better reflect the full nature and context of the experience, they often lack the kind of focus that leads in an obvious direction to specific conclusions and actions that can be taken (O'Brien, 2001; Redmond & Richardson, 2003; Steele & Davies, 2006; Green, 2007a, Manaseri, 2008). The use of a mixed methods approach is a unique strength of this study relative to other published research in this area. This has allowed not only the quantification of the impact of caring for a young child with a life-limiting neurodevelopmental disability on the family, but also helps to understand the nature and origin of the related stress which in turn has identified specific areas for intervention with these families.

The inclusion of both families of young children with life-limiting neurodevelopmental disabilities and the providers of services to this population of children and their families is an additional strength of this study. Although this has been undertaken in previous qualitative studies, these have tended to present the findings as a single overall perspective in which it was impossible to distinguish opinions. The design of this study facilitated the exploration of both convergent and divergent opinion between those who use the service and those who provide it. This is valuable in the context of the differences that exist between to two groups.

However, no single study can capture all that might be learned or known about a given topic. Certainly no study undertaken by a single researcher, within a limited timeframe, can be as comprehensive as the topic might deserve. This study is no exception. The limitations of each individual stage of the study have been discussed in the relevant chapters, however, taken as a whole the study is also subject to several limitations.

Principal amongst the limitations of this study the sample size and the sampling frame from which it was drawn. Although investigating a small population of children and their families, this nonetheless is a relatively small scale study. This, in combination with the lack of any accurate prevalence data on which to base a power analysis, makes it impossible to assess the power of the study. In addition, the sample for this study was drawn from the database of the Jack & Jill Children's Foundation. This represents a group of children and their parents who have services beyond those provided by the traditional health services i.e. a particularly well-supported group of families. This may limit the generalisability of the findings to other families who are not in receipt of these additional services.

Two additional factors limit the generalisability of the findings. The first relates to the self-selection of participants: although all families who met the inclusion criteria for the study were invited to participate, not all wished to do so. Whether these families were significantly different from those included in the study cannot be known. The second issue relates to the homogeneity of the sample. Most of the families who participated in the study were two parent families (married or cohabiting) and there is no diversity in the sample relative to culture or ethnicity.

The study is also limited by its correlational and cross-sectional design. While the design of the study facilitated identifying the relationships between different variables it precluded identification of causal association. Overall the design of the study results in findings, that while comprehensive, represent the experiences of young children with life-limiting neurodevelopmental disabilities and their families, and service providers, at a specific point in time.

9.8 Conclusion

Children with life-limiting neurodevelopmental disability and their families are a unique and vulnerable population. Their complex needs, ongoing requirement for complex and high intensity care, and need for a wide variety of health services and supports differentiate them from other groups of children with disabilities (Bramlett, 2009). Caring for a young child with a life-limiting neurodevelopmental disability at home requires a titanic commitment on the part of the child's family. However despite being confronted with multiple stressors it

was not the child, or the severity of the child's condition, that was the major cause of stress for families in this study. Rather the burden was associated with the sometimes overwhelming, always unpredictable and persistent demands of providing care, which increased family work while simultaneously decreasing social and personal opportunities for mothers. This situation was exacerbated by stigmatising attitudes which lead to social exclusion of the child and family from many normal activities. It was amplified by insufficient, inadequate and bureaucratic services, which instead of ameliorating the stress experienced by families often worked to increase the burden as mothers attempted to navigate the labyrinth of services and entitlements that their child needs.

This chapter has provided a discussion of the key integrated findings of this study. The findings have been mapped to the aims, objectives and research questions outlined in chapter four. The overall strengths and limitations of the study have been presented and discussed.

Chapter 10: Conclusions and Recommendations

“We can succeed only by concert.
It is not, “can any of us imagine better?’ But, “Can we all do better?”

Abraham Lincoln
Annual Message to Congress
Concluding Remarks
December 01, 1862.

10.1 Introduction

This is a challenging time for all health services in Ireland in the context of proposed radical changes to the structure of Irish health systems and the austere fiscal climate in which these changes are taking place (Minister for Health, 2011). Now more than ever it is essential to have services that are designed, and health policy that is developed, on a sound and reliable evidence base. Understanding how services or lack thereof, impact the lives of citizens who experience them and for whom they are ostensibly designed is critical. This study contributes to this understanding in the context of young children with life-limiting neurodevelopmental disabilities and their families.

This concluding chapter presents a synopsis of the key findings of this study and the conclusions drawn from these findings. Recommendations are made, both for clinical practice and for the development of health policy, based upon these conclusions. These recommendations provide a mechanism for improving the quality of life of young children with life-limiting neurodevelopmental disabilities and the quality-of-life of the families who care for them. Directions for future research are presented.

10.2 Summary of Key Findings

It is clear from the findings of this study that while the numbers may be relatively small, the needs of young children with life-limiting neurodevelopmental disabilities are multiple, complex and ongoing, necessitating the provisions of complex medical, nursing and technological skill and care. For the children in this study this care was provided principally by the child’s mother, for which she received little training or recognition. Although the

children in the sample had problems that were significant, both in complexity and number, there was no evidence of an unmet need for specialist palliative care services in this sample of children at this time.

The nature of the child's requirements resulted in an intensive care schedule, which in many cases included the provision of night-time care. Mothers' commitment to the child and the care required was obvious; however this impacted on, and had consequences for, all members of the family, most particularly for the mother as the main provider of hands-on care and management. Family life was characterised by uncertainty and unpredictability, and mothers described episodes of overwhelming burden interspaced by periods of relative stability and routine. In particular crises were associated with discharge from hospital, and oscillations in the child's condition. Where there were other siblings in the family mothers felt guilty about the disproportionate amount of time that needed to be devoted to their most needy child.

For many mothers, rather than a source of support, services designed to support their child were considered an additional cause of stress and burden. Mothers reported services that were difficult to access, insufficient, inconsistent, and fragmented. In addition they found obtaining information about services a consistent challenge. Many of these experiences were supported by the views of the health professionals delivering the services, although service providers appeared to underestimate the way in which services coordinated and integrated to form a cohesive package of care for the child and family. Mothers' additionally reported that communication with, and between, services was problematic at all levels. Despite these ongoing challenges mothers' were fairly satisfied with the services they did manage to obtain, and all reported encountering health professionals who went out of their way to contribute to the welfare of their child and family in a positive way.

When mothers were asked what they needed to improve the care and services they receive the list was headed by easier and more frequent access to the services that their child needs, closely followed by additional practical assistance to help them in their caring role. Alternatively when health professionals were asked what they thought would improve the care and services to this population of children and their families they focused more on improving service related issues.

Despite the involvement of a wide and varied range of service providers in the child's care, there was agreement amongst the group with regards to the overall goals of care for the child. These included some of the principles of a palliative approach to care although the family dimension inherent in this approach was largely lacking. Despite the group agreeing and advancing the concept of home-care for young children with life-limiting neurodevelopmental disabilities, support for the family in the provision of this care was not agreed as a primary goal, nor did it rate in the top-five priorities for improving services to these children and their families.

The findings of this study provide compelling evidence that current services are failing to meet the needs of this population of children and their families. They emphasise the need to urgently address issues related to the inadequate provision of services, and to address issues related to the manner in which the services that are available currently function. There are implications for services at both the practice and policy levels, and while there is an obvious need for more services of all types there is also strong evidence to suggest that more effective multi-agency working, improved service cohesion and streamlining services could also have a major positive impact on families without additional major fiscal commitment. Table 10.1 provides a distilled overview of the key recommendations of this study. These recommendations are discussed in detail in the remainder of the chapter where they are presented separately for improving services to families at the practice and policy levels.

Table 10.1 *Key Recommendations Based upon the Findings of the Study*

<p><u>Key Messages from the Study –</u></p> <ul style="list-style-type: none"> • There are not enough general services available to meet the needs of this population of children and their families. • The services that are available are not sufficiently coordinated or integrated to provide a cohesive and effective package of care. • Service providers lack the “family focus” essential to meeting the needs of families caring for young children with life-limiting neurodevelopmental disabilities. 	
<p><u>Key Recommendations for Practice</u></p> <ol style="list-style-type: none"> 1. Greater multi-agency coordination and shared care planning is essential if care is to be delivered effectively to this population of children and their families. 2. A standardised measurement of need is urgently required against which services are allocated and evaluated. 3. A Family-Nursing orientation to the care of this population of children and their families is essential to address the considerable needs of the family. <p>Responsibility for Implementation All health services personnel who work with the child and family</p> <p>Cost of Implementation Requires improvement and “re-orientation” of current services only. No fiscal commitment required to make these improvements.</p>	<p><u>Key Recommendations for Policy</u></p> <ol style="list-style-type: none"> 1. There is an urgent need for more services of all types. 2. Parents must be involved in the planning and evaluation of services. <p>Responsibility for Implementation Minister for Health DOH&C and Service Managers</p> <p>Cost of Implementation Increasing services to this population of children and their families obviously has fiscal implications, however, this needs to be considered in the context of the impact the lack of services is having on these families.</p> <p>No fiscal commitment required for recommendation 2. This requires only a partnership orientation with parents.</p>

10.3 Key Conclusions and Recommendations for Improving Practice

The findings of this study highlight an obvious need to amend current practice in order to better meet the needs of young children with life-limiting neurodevelopmental disabilities and their families. These necessary changes relate to several aspects of current practice including: greater support for families caring for young children with life-limiting neurodevelopmental disabilities, improving the delivery of services to this population of children and their families, and improving the services themselves.

Although parents identified some positive aspects of current services, principally in terms of attitudes and relationships with professionals and the quality of services received, the overall findings of the study are of services that are difficult to access, insufficient, inconsistent, inequitable, poorly coordinated and integrated, and characterised by bureaucracy and delay. This finding is consistent across both families and service providers. The need to address issues of service provision to this population of children and their families is evident in both phases, and all stages, of this study.

10.3.1 The Transition from Hospital to Home

Although the care required by the young child with a life-limiting neurodevelopmental disability was consistently burdensome, there were situations and episodes that were particularly traumatic and overwhelming for mothers. One of the greatest challenges mothers identified was in relation to the initial discharge from hospital with their infant. Many described being overwhelmed by the intensive care requirements of the child and lack of community based support services. In some instances mothers and infants were discharged without referral to community services and it was up to the mother herself to try to organise this. This appeared to be an incredibly difficult time for mothers as they grappled with the complex needs of their child, with all mothers identifying this as a time of particular vulnerability and crisis. The complexity of the learning curve for parents at this stage in their journey has been described in previous studies related to children to children with life-limiting conditions and complex health care needs (O'Brien, 2001; Alexander *et al*, 2002; Redmond & Richardson, 2003; Quinn *et al*, 2005; Steele & Davies, 2006).

The opinion of service providers in this study was that hospital based services are not sufficiently aware of the services available to these children and their families in the community. This may have contributed to the deficit in community based services that mothers experienced on discharge, however, it does not account for the fact that many mothers did not feel sufficiently prepared to take their child home in the first place. Careful planning and preparation at a number of levels is vital for the critical transition of the child and family from hospital to the community. Lewis and Noyes (2007) suggest that the discharge of a child with complex healthcare needs is a process that needs to be planned across agencies, with all individuals who are involved in that process understanding and contributing to the decisions made. Harrigan *et al* (2002) used a review of the literature related to medically fragile children and their families to develop a framework for improving the quality of care delivered. Amongst the suggestions is that the training for discharge must be meticulous, extensive and as real-world as possible, and that families should be well connected to available community services before being discharged from hospital. Similarly Price and Thomas (2007) propose the need for a comprehensive and robust training programme for parents of children with complex needs prior to discharge. This would appear to be particularly important for mothers of children who are technology dependent to ensure that they are provided with the skills, knowledge and confidence to care for their child at home.

Recommendations

There should be considered, planned and meticulous discharge preparation for young children with life-limiting neurodevelopmental disabilities and their mothers. This should be initiated well in advance of the proposed discharge from hospital so that mothers are appropriately trained in the management of their child's care, and confident with using any technology that the child requires.

- Discharge planning must involve both hospital and community based services, in particular Public Health Nurses and General Practitioners as the primary providers of care in the community. Such a collaborative system of discharge planning would ensure that all children and their families are connected to the appropriate community based services before discharge.
- A directory of appropriate locally available services should be available to all mothers. This would alleviate the consistent difficulty that mothers experience with

accessing information about services, and is particularly important since community based services are agreed to be inequitable and not uniformly distributed.

10.3.2 Adopting a Family-Nursing Approach to Care

The stressors facing a family caring for a young child with a life-limiting neurodevelopmental disability are complex and multiple. Ongoing care provision has consequences for all family members, and all aspects of family life. The findings of this study underscore the need for a family nursing approach to the care of young children with life-limiting neurodevelopmental disabilities and their families.

The concept of “family” has been variously defined in health-related literature, with definitions differing depending upon the theoretical orientation of the definer (Freidman *et al*, 2003). Subsequently family nursing can be conceptualised in a variety of ways which in turn will influence the manner in which it is practiced. Although Neabel *et al* (2000) suggest that the purpose of family nursing is to enhance the promotion of family strengths and coping skills, which may help to maintain the family’s health, in reality the degree of family-centeredness is dependent to a large extent upon the philosophy of the system in which it is practiced. The concept of family nursing is not widely used in current healthcare practice or policy although recognition of the “patient and family” or “child and family” is found in many policy documents and nursing curricula. Family-centred or family-focused care is the more frequently used term (DOH&C 2004a DOH&C 2000, DOH&C 1999), which while acknowledging the importance of the child’s family, and advocating health services work “in partnership with families” (DOH&C 1999, DOHC 2004b, HSE 2005), does not necessarily imply a readiness to consider not only the needs of other family members but the needs of the family unit as a whole. Nor does it imply the inclusion of families in the planning, implementation or delivery of care and services.

More recent health policy and legislation, particularly in the context of disability services, has further narrowed the focus of care with an emphasis on “person-centred care” (DOH&C 2007, DOH&C 2006, Government of Ireland 2005). At a time when all health policy is shifting the focus of care to the community wherever possible (DOH&C 2008a, DOH&C 2008b, DOH&C 2007, DOH&C 2001a, DOH&C 2001b), particularly in the context of

children with life-limiting conditions (DOH&C, 2010) the case for addressing the needs of families, in this context the primary carers, seems obvious. It must also be acknowledged that different families react differently to the demands placed upon them. The considerable morbidity experienced by the families of young children with life-limiting neurodevelopmental disabilities emphasises the importance of addressing the family dimension of the care delivered. Adopting a family-nursing approach facilitates a move to a view of the family as a unit of care rather than simply a context for care. The expert panel in this study agree that this is not currently the perspective of service providers to these children and their families.

Mothers in this study provided valuable, highly skilled, and complex care to their children; this expertise needs to be acknowledged by all health care providers by adopting a partnership relationship with mothers. Although partnership is at the core of all Irish child and health care policy it was not evident in the findings of this study, particularly in relation to hospital based services, and there appeared to be discrepant perceptions between mothers and service providers of what is involved. Rather than a genuine partnership approach mothers in this study reported what de Lima *et al* (2001:562) described as a “Marxist division of labour”: whereby mothers perform the ‘manual’ work in caring for the child, while health professionals carry out the ‘intellectual’ work. Mothers in this study were not in control of any aspects of the planning, goal-setting, decision making or coordination of care for their child.

Recommendations

- A family nursing approach is required in the delivery of care and services to young children with life-limiting neurodevelopmental disabilities and their families. This approach would facilitate the identification of the needs of each member of the family and of the family as a functional unit.
- Service providers need to adopt a genuine partnership model with parents in which mothers’ expertise in relation to the care of their child is appropriately acknowledged and valued.

10.3.3 Planned and Ongoing Assessment of the Needs of the Child and Family

This study highlights the multidimensional and complicated nature of need with regards to children with life-limiting neurodevelopmental disabilities and their families. The findings suggest that considerable morbidity may be experienced by all family members in particular the mother as the main provider of care, who placed their own needs second to those of all other family members. Family needs are subject to change, and in this study stressors were perceived more acutely in association with changes in the child's condition, episodes of hospitalisation, or events necessitating unanticipated changes to the established family routine. Such findings are not new (O'Brien, 2001; Alexander *et al*, 2002; Redmond & Richardson, 2003; Quinn *et al*, 2005; Steele & Davies, 2006), however they do point to the importance of frequent and planned re-assessment of the family's needs. It should also be acknowledged that, despite many commonalities, each family has a unique experience, including unique strengths and adaptive abilities. These should also be considered a focus for intervention with families.

Recommendations

- There should be comprehensive assessment of the needs of the child, the needs of each member of the family, and of the family as a whole. This must be subject to ongoing review and not occur only in crisis situations. It is particularly important in association with episodes known to increase the stress experienced by the family.
- In addition to a care-plan for the child, a family care-plan which addresses the needs of the family should be developed. Best practice suggests that such a care-plan should be genuinely multi-disciplinary and have a multi-agency focus (ACT, 2003c). In addition to addressing the needs of the family and improving family quality-of-life such a care-plan would foster effective multi-agency working and collaboration.
- This assessment should include the unique strengths and capabilities of each family.

10.3.4 Addressing Insufficient Services

Providing care and support to young children with life-limiting neurodevelopmental disabilities can be both physically and mentally exhausting for mothers. Families need appropriate support to sustain and support them in their caring role, not only in times of crisis

but on a day-to-day basis. Parents need services and supports that are accessible, timely, and useful. They do not need to be wasting time and energy that would be better spent on their child and family battling for services and equipment that are more trouble than they are worth.

There were many areas of insufficient services identified in this study. Mothers reported, and health care providers acknowledged, that currently services are simply not sufficient to meet the needs of these children and their families. The heavy reliance on charity services to address this gap was acknowledged by both by mothers and service providers. Parents identified the negative impact this had on both the child and the family, while service providers reported that in order to address this issue services to children and families needed to be rationed. Parents expressed a need for more therapeutic and support services of all types, and an urgent need for respite services and support services including practical support in the home.

Consistent with the recent review *Respite Services for Children with Life-Limiting Conditions and their Families* (Irish Hospice Foundation & Children's Sunshine Home, 2011) the need for more, and better, in-home respite for mothers was obvious in this study. This was not an uncomplicated issue however, and several challenges were identified in association with this issue. These were principally related to the complexity of the child's care which resulted in a difficulty obtaining qualified carers, and the lack of available qualified carers resulted in the provision of respite when it was available rather than when it best met the needs of the family. In addition lack of night-time respite was particularly problematic for families whose child required round-the-clock care. It may be that alternative models of service provision need to be considered to address this issue in a genuinely meaningful way. Precious and limited financial resources may be better spent in a consumer-based model of respite provision, whereby mothers were provided with the finances to purchase their own respite (through a nursing agency or other organisation), rather than the current service based model. Such a model could also be applied to the provision of practical support services such as home-help or home-support workers, whereby mothers could negotiate directly with the individual the hours that would best suit the child and family and the role that the support worker would perform.

Despite the high levels of emotional stress amongst the mothers in this study, psychological support services for these parents, and siblings, are currently insufficient. The lack of such services was identified as a deficit by the mothers in this study, and confirmed by the service providers. There are no studies exploring the longer-term consequences of the emotional burden of caring for a child with a life-limiting neurodevelopmental disability. However it is possible that failing to intervene appropriately at an early stage may result in the need for a greater number of resources in the long term.

Recommendations

- There is an urgent need for more services of all types. This includes therapeutic and in-home support services. Given the difficulties associated with obtaining qualified carers alternative models of service provision may need to be explored.
- Psychological support services for parents and siblings should be developed. However, the lack of evidence to support the efficacy of any particular model or intervention to address this issue would suggest that this can only be developed after appropriate and relevant research has been undertaken.

10.3.5 Addressing Service Deficiencies

In addition to insufficient or absent services, parents also identified deficiencies in the services that are available. In their experience of using services parents' reported difficulties with information and access, coordination and integration of services, and bureaucracy and delays. Many of these difficulties were also openly acknowledged by service providers. Mothers described these challenges as an additional and significant source of stress and burden. Hewitt-Taylor (2005) suggests that, particularly in the home environment, the lack of coordination between services can overwhelm parents. In addition, Glendinning & Kirk (2000) suggest that where communication is poor, parents may feel that they are the only liaison between services and individuals which does little to add to their confidence in the service.

The need for a designated key-worker for every family caring for a child with a life-limiting neurodevelopmental disability is apparent in all stages of this study. It was identified as a deficit by both mothers and health service providers, has previously been identified as a

requirement by Quinn *et al* (2005), and forms part of the current policy for improving care for children with life-limiting conditions (DOH&C, 2010). Mothers who had a key-worker in this study identified it as a resource that worked well for them. The advantages of a designated key-worker for families are multitude. The provision of a designated key-worker for all families has the potential to address many of the deficits and on-going challenges that mothers experience on a daily basis including: the provision of information about services and entitlements, improving access to services, coordinating the services the child receives, and acting as a support and resource for parents. It would also address the problems that mothers report in relation to continuity of care for the child and family, and may improve family assessment if the key-worker was available to the family over the course of the child's and family's journey. A designated key-worker may also reduce the challenges that mothers' reported in relation to the communication difficulties they experienced with the health professionals and service agencies involved in the care of their child.

Recommendation

- Every family caring for a young child with a life-limiting neurodevelopmental disability should have a named designated key-worker assigned to work with them on an ongoing basis.

10.4 Key Policy Recommendations

While some of the difficulties and challenges experienced by parents in this study can be addressed at the local level of service provision there are others that require intervention at a higher policy level.

10.4.1 Funding Issues

In its policy document *Working for Children and Families: Exploring Good Practice* (DOH&C, 2004) the Department of Health and Children propose that services to children and their families must have adequate resources to meet their objectives, and that in order to be effective services need to be responsive to need. Neither of these aspirations appears to have been achieved in the context of the children and families in this study where consistently parents and service providers report that services are underfunded and not sufficiently resourced. In this context the issue of funding and resourcing services to this group of

children and their families needs to be reconsidered. While some of the findings of this study highlight the need for better ways of working, and as such have minimal fiscal implications, it is impossible to avoid the fact that that parents experiences of using services, and health professionals experiences of providing services, suggest that there are simply not enough services available to meet the needs of this population of children and their families. No amount of creative adaptation of current services or better ways of working can alter this deficit without additional funding.

Obviously funding of services is contingent upon the fiscal resources available, and certainly in the current climate these are severely limited. However, failure to sufficiently fund services to this vulnerable group of children and their families at this stage may in fact have implications for service usage in the future, particularly if, as mothers suggest lack of services negatively impacts on the child's condition and causes considerable stress for families. In addition it likely contributes to the prevalence of psychological morbidity experienced by the mothers in this study. The acknowledged dependence on charity services to meet the needs of this group of children and their families needs to be questioned. It is possible that the financial resources available to this charity will be negatively impacted by the same climate of financial austerity that has resulted in the scaling back of other health services. The consequences of this for this population of children and their families would be severe.

Recommendation

- Funding of services needs to be reconsidered in the context of the impact the lack of services is having on this group of children and their families.

10.4.2 Issues of Inequality

The limited services that are available to children with life-limiting conditions and their families are neither equitably nor fairly distributed. This is the perception of service providers, and the experience of parents, both of whom report that the services currently available depend to a large extent upon where the child and family live. This represents a fundamental ethical and moral issue at the policy level that needs to be addressed. There is

little point in having additional services if the benefits that such services bring are not universally available to all children and families who need them.

It is possible that a number of factors contribute to this situation: the relatively small numbers of children and families involved; the various combinations of statutory, voluntary and charity service provision; the lack of any real coordination of services at a national level; and the absence of any agreed standards against which to measure service provision. Regardless of the cause, this is a service issue that needs to be addressed and services to these children and their families should be apportioned on the basis of the individual needs of each child and family.

In order to achieve this an objective, standardised measure of need is required. This requirement for such a measure is particularly obvious in the allocation of in-home support services such as respite and home support hours. Currently these services are allocated on an arbitrary basis, dependent upon what services are locally available, and what funds are locally available. Mothers report in-home support services that are erratic and can be reduced or withdrawn without notice or explanation. While a standardised measure of need will not improve the limited services that are available it may provide the basis for a more equitable distribution of these limited resources at a local level. It could also be used to identify deficits, or inequalities, at a national level and therefore form the basis of additional resource allocation.

Recommendation

- A standardised, objective measure of need should be developed, and used as the basis of resource allocation, so that every family caring for young child with a life-limiting neurodevelopmental disability would have equal access to the support services necessary to sustain them in their caring role.
- National or regional coordinators of care for children with life-limiting conditions should be appointed to ensure equitable and uniform provision of services that are appropriate to the needs of each individual child and family.

10.4.3 Involving Parents in Service Planning and Evaluation

Gardner *et al* (2001) suggests that defining outcomes intended for children and their families, and specifying the services to be provided to meet these outcomes, is the foremost purpose of any service delivery system. The findings of this study support previous research (Hunt, Elston and Galloway, 2003) in suggesting discrepant views between parents and service providers in relation to how services work to meet the needs of families, and in relation to which services are required to better meet these needs. Meeting the needs of these children will continue to challenge multiagency working as more vulnerable children with life-limiting neurodevelopmental disabilities continue to survive. In the context of limited resources, and to ensure services are effective in meeting the needs of those they were designed to serve, parents should be involved in the planning and evaluation aspect of service provision. Whitton *et al* (2008) suggest that this is particularly useful in identifying local solutions to any structural inter-agency barriers.

Recommendation

- A strategy should be developed to involve parents in service planning and evaluation.

The previous sections of this chapter have recommended changes that are required to improve the provision of services to children with life-limiting neurodevelopmental disabilities and their families based upon the findings of this study. Although some of these recommendations require additional fiscal resources others simply require better methods of multi-agency working and a different service orientation, neither of which have major cost implications. Some of these findings of this study are not new and support previous research undertaken with similar populations of children and their families in a variety of international service contexts which suggest that the problems experienced by these families are universal. However, this study has identified that while there are many aspects of family stress that service providers cannot change, they do have ongoing opportunities to foster parents' resilience by providing them with the support they require to continue to care for their child at home. In this respect the study has identified a specific focus for intervention with these families. Understanding the distinctive aspects of stress related to caring for a child with a life-limiting neurodevelopmental disability, such as the considerable burden of care, living with chronic uncertainty and change, and mothers ongoing struggles for services and for

validation of their expertise can offer insights about experiences that occur beyond a professional's normal gaze or personal interactions with a family.

This study has also identified the unique opportunity health professionals have to mediate many of the avoidable stresses that are a part of these families' journeys. If, as the parents in this study suggest, professionals in multidisciplinary systems provide comprehensive and coordinated services with both competence and compassion, the reality of parenting a child with a life-limiting neurodevelopmental disability would not change, but the experience would include fewer avoidable stresses, and meeting their child's needs would be considerable less challenging for parents.

Although every effort was made to conduct this study in as rigorous a manner as possible the study was not without the limitations previously discussed. Future research can address these limitations, and expand the findings of the study and their implications for practice. The final section of this chapter makes recommendations for future research with this population of children and their families.

10.5 Directions for Future Research

Liben, Papadatou and Wolfe (2008) suggest that there are many more questions than answers about how best to care for children who will probably die before adulthood, and while this study had attempted to address some of these issues there are critical areas yet to be explored. There is relatively little known about this population of children and their families over the course of their journey. Studies exploring young children with life-limiting conditions and their families have tended to be either descriptive qualitative designs, or cross-sectional survey designs. More longitudinal research is needed to explore the needs of these children and their families over the course of the caring trajectory.

In particular research is needed to explore the morbidity experienced by the children and its management over time. There is also a need to explore the end-of-life care trajectory for these children. This is particularly important in order to establish the need for, and potential benefits of, providing specialist palliative care services to this population of children and their families.

There is also a need to explore the long-term consequences of care provision on families. While there is a considerable volume of research supporting the high levels of stress experienced by families caring for children with life-limiting conditions or complex needs this is cross-sectional in nature, subsequently the consequences of this chronic stress for the family in the longer-term is not known. In addition, there is little research exploring the experiences of siblings of young children with life-limiting neurodevelopmental disabilities, or indeed the siblings of children with any life-limiting conditions. This is an area that also needs to be addressed.

There is a considerable volume of research measuring, or describing, the psychological distress and emotional strain experienced by parents of children with life-limiting conditions. However, research on appropriate interventions to alleviate this is largely lacking. Although difficult to design (the small size of the population makes conducting randomised trials difficult) there is an urgent need for additional research exploring the efficacy of different forms of intervention to support parents in their care-giving role.

There has been concern that, within the growing interest in parents' perspectives in paediatric palliative care, the research does not equally reflect the experiences of mothers and fathers (MacDonald *et al*, 2010). Additionally, in an analysis of current literature on the experience parenting children with health problems, Pelchat *et al* (2007) conclude that fathers and mothers experience this ordeal differently. There is certainly a lack of research studies exploring fathers' perspectives and experiences in the context of young children with life-limiting neurodevelopmental disabilities. This gap needs to be addressed.

10.6 Conclusion

Current national health and social policy (DOH&C, 2010) is consistent with international recommendations in proposing home as the optimum place of care for all children with life-limiting conditions regardless of the complexity or nature of the child's care (Dangel, 2002; Radbruch & Payne, 2009). However, caring for a child with a life-limiting neurodevelopmental disability at home is an extraordinary challenge and takes remarkable commitment on the part of the caregivers, principally mothers. The level and complexity of

the care required is prodigious, and families need to have support from a cohesive family intervention plan to buffer the stresses that constantly arise.

Research literature would suggest that the experience of stress is a shared and ubiquitous experience for all parents of a child who has a life-limiting or complex medical condition or severe disability. Unless cures are found for all childhood life-limiting conditions and complex disabilities this inexorable journey of stress, that becomes an inevitable part of a parent's experience of caring for their child, cannot be changed. Service providers cannot alter the devastating fact that a child has a life-limiting neurodevelopmental disability, nor can they change the fact that parents deeply love and worry about their children. These sources of stress are unavoidable. However parents also report significant stress associated with trying to obtain both the care and services that their child needs and the resources required to sustain them in their caring role. It is possible for many of these avoidable and unnecessary stresses to be mediated or even eliminated entirely for families. So while health professionals cannot change the stressful reality of a child's condition, they can have a positive impact on how that reality is experienced by providing the support and services necessary to assist parents in their caring role.

Services providing care to this population of children and their families face many complex issues including issues related to funding, communication, and coordination and integration. What is required is an integrated set of services that optimise the use of limited resources and coordinate to address the unique needs of each child and family. Creative adaptation of current services, to provide high quality, coordinated and comprehensive care to this vulnerable population of children and their families may better meet their considerable needs that the introduction of additional and costly specialist services.

This chapter has provided a summary of the key findings of this study. These findings achieve the overall aim of the study which was to provide a detailed and reliable evidence base related to the current health system as it pertains to children with life-limiting neurodevelopmental disabilities and their families. Based on these findings recommendations for practice and policy were made with a view to improving services to this population of vulnerable children and their families and subsequently improving quality-of-life and family outcomes for all involved.

