

# **An exploration of the experience of caring for children with non-malignant life-limiting conditions including, accessing services and decision-making surrounding the child's care**

**A thesis presented to the School of Nursing and Human Sciences,  
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Doctor of Philosophy**

**Fiona Hurley**

MSc. Nursing, BSc Nursing (Hons), Higher Diploma in Nursing Studies (Sick Children's Nursing) RGN, RCN, RNT

**Academic supervisors:**

Dr. Gemma Kiernan

School of Nursing and Human Sciences, Dublin City University

Professor Jayne Price

School of Nursing, Faculty of Health Social Care and Education, Kingston University and St George's University London.

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## Declaration

I hereby certify that this material, which I now submit for assessment on the programme of study leading to the award of degree of Doctor of Philosophy is entirely my own work, that I have exercised reasonable care to ensure that the work is original, and does not to the best of my knowledge breach any law of copyright, and has not been taken from the work of others save and to the extent that such work has been cited and acknowledged within the text of my work.

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(Candidate) ID No.: 13211391

Date: 20<sup>th</sup> of November 2019

## **Dedication**

### **Dedicated to:**

The most inspiring infants, children, young people and parents I have ever met. It was a privilege to have had the wonderful opportunity to spend time with you.

## Acknowledgements

I would like to extend my sincere gratitude to all who have helped bring the study to its conclusion.

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# Operational Definitions

## **Child**

A child is defined as a young person aged up to their nineteenth birthday (Together for Short Lives, 2018).

## **Children's hospice services**

Children's hospice services provide palliative care for children and young people with life-limiting conditions and their families. Delivered by a multi-disciplinary team and in partnership with other agencies, children's hospice services take a holistic approach to care, aiming to meet the needs of both child and family - physical, emotional, social and spiritual - through a range of services (Together for Short Lives, 2018).

## **Children's palliative care**

Palliative care for children is 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 and support for the family. It includes the management of distressing symptoms, provision of short breaks and care through death and bereavement' (ACT 2009, p.7; Together for Short Lives, 2018).

## **End-of-life care**

End-of-life care is care that helps all those with advanced, progressive, incurable illness, to live as well as possible until they die. It focuses on preparing for an anticipated death and managing the end stage of a terminal medical condition. This includes care during and around the time of death, and immediately afterwards. It enables the supportive and palliative care needs of both child/young person and the family to be identified and met throughout the last phase of life and into bereavement. It includes management of pain and other symptoms and provision of psychological, social, spiritual and practical support and support for the family into bereavement (Together for Short Lives 2013; Together for Short Lives, 2018).

## **Family**

The term 'family' includes parents, other family members involved in the care of the young person, or other carers who are acting in the role of parents. Family includes informal carers and all those who matter to the child/young person (Together for Short Lives, 2018).

## **Ireland**

Ireland is used to describe the twenty-six counties in the Republic of Ireland.

## **Life-limiting**

A life-limiting condition in a child is described as any condition from which there is no reasonable hope of cure and from which the child or young adult will die (ACT, 2009; Together for Short Lives, 2015; Together for Short Lives, 2018). While the majority of children with such a condition are unlikely to live beyond eighteen years, some of those diagnosed in childhood can survive unexpectedly into early adulthood. Life-limiting conditions in children can be diagnosed either prior to birth or during childhood, with the expectation that the condition will lead to premature death.

## **Life-threatening**

Life-threatening conditions are those for which curative treatment may be feasible but can fail, such as cancer. Children in long-term remission or following successful curative treatment are not included (Together for Short Lives, 2018).

## **Northern Ireland**

When reference is made to the North/Northern Ireland it is used to describe the six counties in Northern Ireland.

## **Parents**

The term 'parents' is used to describe any carer for a child whether that is a married or unmarried couple, a single parent, guardian or foster parent (Together for Short Lives, 2018).

## Abstract

Children with non-malignant life-limiting conditions (NMLLCs) constitute the largest proportion of children requiring palliative care. Partly due to technological advances, growing numbers of these children are experiencing improved survival. Care is mostly provided at home by the family, with significant input from a range of health and social care providers at different points in the life trajectory. Little is known about parental experiences of caring for a child with a NMLLC including their experiences of accessing services and decision-making. Similarly, a dearth of information exists regarding the experiences of healthcare professionals caring for this cohort of children. This study aimed to explore the experience of caring for children with NMLLCs including, the experience of accessing services and decision-making. Participants were twenty-three parents (eighteen mothers and five fathers) and twelve healthcare professionals' recruited from five organisations with a wide geographical remit. A concurrent mixed methods design was adopted. Qualitative data were collected from parents and healthcare professionals through semi-structured interviews. Two standardised measures and a demographic questionnaire collected quantitative data from parents. The measures were the Decisional Conflict Scale (DCS) and the Measure of Processes of Care-20 Scale (MPOC-20). The qualitative data took precedence within this concurrent embedded mixed methods design. Three key themes which emerged from the interviews with the parents were 'Starting out in haziness', 'Managing an unexpected life' and 'The ship is going to go down'. Parents reported a generally low level of decisional conflict in the study. In addition, parents reported a generally positive perception of family-centred care provided. Three key themes which emerged from the interviews with the healthcare professionals were 'Being there', 'Being focused' and 'Being strong'. Although commonalities across parents' and professionals' accounts were noted, key differences were also apparent. Parents and healthcare professionals equally demonstrated an extraordinary level of commitment to the child, focusing on providing expert care. Whilst aspects of good service provision were highlighted, both participant groupings identified that overall current services seemed under-resourced and insufficient to meet child and family needs. However, the perceived need to fight for services and the need to make complex decisions were more apparent in the parents' accounts. This study is unique as no other study has explored the palliative care needs of children with NMLLCs and their families in an Irish context.

# Chapter 1: Introduction

## 1.0 Introduction

This thesis explores the experience of caring for children with NMLLCs on the island of Ireland both (north and south) including, the experience of accessing services and decision-making surrounding the child's care. An All-Ireland approach was chosen in order to provide a broad perspective on the range of experiences across two healthcare systems. In this introductory chapter the author sets the context for the study. To commence, matters pertaining to the definition of 'life-limiting' conditions in children are discussed and the pertinent concepts relating to the study are explored. The concept of palliative care as it pertains to children is introduced. Subsequently the classifications of life-limiting conditions in children are reported and the prevalence presented. Palliative care service provision for children internationally and in the United Kingdom and Ireland is discussed. To conclude, the thesis conspectus outlines the breakdown of each chapter.

## 1.1 Matters pertaining to the definition of life-limiting conditions in children

Although the terms 'life-limiting' and 'life-threatening' are common and often interchangeably used to describe the spectrum of children that need palliative care, there has been considerable debate in the literature about the ambiguity of these terms. In the international context, in particular, the terms 'life-limiting' and 'life-threatening' tend to be used interchangeably with no differentiation made between them. However, there are differences between the two terms. Children are usually said to have a 'life-limiting' condition where there is no reasonable hope of cure and a 'life-threatening' condition where the possibility of cure exists (Together for Short Lives, 2018). In practice, the distinction is often arbitrary since an individual child may oscillate between the two especially during acute exacerbations of the illness, for example children with cystic fibrosis (Rodriguez and King, 2014). There is a further complexity in that the term 'life-threatening' is the more inclusive term of the two as it encapsulates all children who may die, but it is also used to describe a very precise point in the illness trajectory when a child has a 'life-threatening episode' and there is an immediate threat to death. Even the term 'life-limiting' is not unambiguous, since for some it implies limitation of ability rather than lifespan (Noyes et al., 2013). The term 'life-shortening' is also entering the lexicon of language in Children's Palliative Care (CPC) (Spicer et al., 2015; Together for Short Lives, 2018). Understandably clear and unequivocal language is essential in CPC. For the purpose of this study, the term 'life-limiting' will be used as it is the most commonly used term in Ireland both (north and south) to describe the range of children who need palliative

care. It is defined as conditions from which there is no reasonable hope of cure and from which children and young people will die (Together for Short Lives, 2018).

## **1.2 The concept of experience(s), access and decision-making**

The importance of exploring experiences, expectations and satisfaction with healthcare has long been recognised (Bowling et al., 2012; Grocott and McSherry, 2018). In addition, numerous conceptual models and frameworks offer a wide array of definitions and insights into framing experiences in healthcare (Browall et al., 2013; Katigbak et al., 2015). Entwistle et al. (2012) identified the range of experiences of healthcare delivery that matter to service users and produced a conceptual map to facilitate consideration of why they matter. The conceptual map presents a rich array of experiences, including healthcare relationships (beyond communication) and their implications for people's valued capabilities (e.g. to feel respected and experience reciprocity). The conceptual map links healthcare delivery to what people are enabled (or not) to feel, be and do and highlights the broad implications of the social dynamics of healthcare delivery. Experiences are labelled from a patient's/client's perspective, rendering the importance of responsiveness to individuals axiomatic (Entwistle et al., 2012). Significantly the subjective experience is emphasised within each of the conceptual models, frameworks and maps. It is within these conceptual models, frameworks and maps that experiences are considered.

Access is central to the performance of healthcare systems around the world. However, access to healthcare remains a complex notion as exemplified in the variety of interpretations of the concept across authors (Cylus and Papanicolas, 2015; Evans, Hsu and Boerma, 2013; Hall et al., 2008; Nasiripour, Mahmoudi and Raeissi, 2011; Schneider and Devitt, 2017). Levesque, Harris and Russell (2013) view access as the opportunity to identify healthcare needs, to seek healthcare services, to reach, to obtain or use healthcare services, and to actually have a need for services fulfilled. They conceptualise five dimensions of accessibility: 1) Approachability; 2) Acceptability; 3) Availability and accommodation; 4) Affordability; 5) Appropriateness. Access, in this study, ultimately refers to the ability of families to obtain appropriate and necessary healthcare services for the child and the family unit as a whole. This definition was chosen as it introduces a concept of access to health care describing broad dimensions and determinants that integrate demand and supply-side-factors and enables operationalisation of access to health care all along the process of obtaining care and benefitting from services. It has been informed by the preceding concept of access (Levesque, Harris and Russell, 2013).

Decision-making refers to selecting a course of action from two or more alternatives. It is initiated to achieve a specific objective or to solve a specific problem (Akrani, 2010). The literature suggests that a number of significant decisions may need to be considered for children with life-limiting conditions over the course of their life (Carroll et al., 2012; Tamburro et al., 2011; Wright et al., 2009). Such decisions are complex and influenced by more than simple exchange of information (Carroll et al., 2012). Within this study, decision-making will refer to choices made regarding current treatment and care options or future care plans and will focus not only on the decisions parents make but on the process of making them. Past decisions will also be considered. The healthcare professionals' views relating to decision-making will be likewise deliberated.

### **1.3 Palliative care concepts**

The concept of palliative care is derived from the Latin word '*palliate*' which means to "conceal or alleviate symptoms without curing" (Romesberg, 2007, p.111). This focus on comfort and supportive care in contrast to continued aggressive intervention forms the basis of palliative care (Spicer et al., 2015). It is a philosophy of care that evolved from the hospice philosophy to address the gaps in care for seriously ill and dying patients (Knapp, 2009). Dame Cicely Saunders, more than anybody else, was responsible for establishing the discipline and culture of palliative care (Campion, 2011). By introducing effective pain management and insisting on rigorous scientific research in end-of-life care, she succeeded in abolishing the prevailing ethic that dying patients should be cured, and failure to do so was a sign of defeat. She also introduced the idea of 'total pain,' a concept which includes the physical, emotional, social and spiritual dimensions of distress- all of which had to be addressed if the dying patient was to experience a good death (Campion, 2011; Dobson, 2017). Dame Cicely Saunders established the first modern adult hospice (Baines, 2013). St Christopher's Hospice opened in South West London in 1967. This hospice was dedicated to research, teaching and expert pain and symptom control. CPC is much more recent in development (Hain and Wallace, 2008; Noyes et al., 2013). Helen House, the world's first children's hospice, opened in Oxford, in 1982. Its development was envisioned by the founder Sr. Frances Dominica and the parents of a little girl with a life-limiting condition, called Helen. Helen lived at home with her family, but required twenty-four hour care, and as a result her parents recognised the need for respite care in a suitable environment. The initial years of Helen's illness and the challenges and experiences that her parents encountered are important, because they became a key influence in shaping the philosophy of Helen House as 'a home away from home' (Worswick, 2000).

However, finding concise language to describe the breadth and complexity of CPC continues to pose an ongoing challenge. CPC is still frequently linked to end-of-life care, with the belief in minimising treatment interventions (Rallison and Raffin-Bouchal, 2013; Together for Short Lives, 2018). This is particularly worrying for this population with lengthy illness trajectories, where there is a need for intense care for the child and family from the point of diagnosis or recognition of a life-limiting condition for the duration of the child's life and beyond. End-of-life care is only one component of CPC. 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 conditions and their families. It is defined as "an active and total approach to care, which embraces physical, emotional, social and spiritual elements" (ACT 2009, p.7). It focuses on enhancement of quality of life for the child and support for the family (Zimmernann et al., 2016). It includes the management of distressing symptoms, provision of respite care and care through death and bereavement. It is an approach to care which can be used exclusively or in conjunction with curative treatments (Together for Short Lives, 2013; Together for Short Lives, 2018). It is not about dying, but rather about comfort and enhanced quality of life for the child (Hill and Coyne, 2012). Thus, CPC is focused on ensuring the best possible quality of life for children and their families whose illness makes it likely that they will not live to become adults (Liben, Papadatou and Wolfe, 2008). Clearly, the definition is broad. Importantly, it includes the care of all children with life-limiting conditions.

While the principles of palliative care for children and adults are comparable, there are many ways in which CPC is unique (Bergstrasser, 2018). These include the diverse illness conditions and the rarity and unpredictability of many childhood life-limiting conditions (ACT, 2009; DoHC, 2009; Hain et al., 2013; Spathis et al., 2012). Many of the conditions are genetic and thus may affect more than one child in the family (Benini et al., 2008; Together for Short Lives, 2018). Even children with the same condition can have different needs and illness trajectories, as seen in recent studies looking at survival rates for children with Duchenne muscular dystrophy (Kimura et al., 2013; Pegoraro et al., 2011). Thus, each child requiring palliative care is unique. Their needs are influenced by their age, cognition and level of support they have in their lives. Parents are generally more involved as direct caregivers and decision-makers in CPC than in the adult setting (Koch and Jones, 2018).

Furthermore, parents are often experts in their child's care and assume responsibility for carrying out a range of technical healthcare procedures (Spiers and Beresford, 2017).

Palliative care for children is now recognised as a distinct specialty focused on providing holistic and integrated care across the care trajectory to enhance the quality of life of children and young people

with a life-limiting condition (Collins et al., 2016; Knapp et al., 2011; Siden, 2018). Palliative care for children adopts the philosophy of a family-centred approach. Central to this approach is the belief that the child is part of a family system and therefore both the child and his or her family are the unit of care (Classen, 2012; Knapp and Contro, 2009; Knapp et al., 2010). Within children's healthcare the concept of family-centred care has been the preferred philosophy of care for many decades. However, family-centred care is seen as 'at a crossroads today' because fundamental misunderstandings persist about what it is (Dennis et al., 2017; Kuo et al., 2012). There is confusion regarding core principles and rival terms and uncertainty with regard to how to implement it. Although family-centred care is a complex concept that is poorly delineated (Mikkelsen and Frederiksen, 2011; Smith, 2018), it is often the preferred approach to structure provision of children's care. Over the past decade, the concept of child-centred care has been gaining support as an alternative approach for care (Carter et al., 2014; Smith and Coleman, 2009). Ford et al. (2018) define child-centred care as any approach to or philosophy of care which is characterised by positioning the child at the centre of care (whilst acknowledging the importance of parents/carers and family). This core definition is linked to/underpinned by one or more of the following characteristics: a consideration of childhood and the developing child; an acknowledgement of children as individuals and the promotion of opportunities for children to be engaged actively within their care (Ford et al., 2018). A child-centred approach has been embraced as a philosophy in CPC particularly in relation to the transition to adult services for children and young people with palliative care needs (Doug et al., 2011; Kerr et al., 2017). However the meanings of this concept are not fully described in current literature (Söderbäck, Coyne and Harder, 2011). Further work to explicate the tenets underpinning child-centred care is currently in progress with collaboration of many academics and nurse scientists from paediatrics and child health worldwide (Shields, 2018).

Palliative care for children has emerged in response to the increased prevalence and recognition of children who have been diagnosed with a life-limiting illness or condition (Noyes et al., 2013; Quinn and Bailey, 2011). Although still a relatively small number, this group of children and their families require intense and sustained support throughout the child's life as well as for several years beyond (Price, 2012). Indeed professional duties and responsibilities do not cease when a child dies as families require compassionate support at this stage of the care trajectory (Jones, Contro and Koch, 2014; Zimmermann et al., 2016). The quality of care at end-of-life and after the child's death can have a major impact on the family's grieving and how they live the rest of their lives (Snaman et al., 2017). It is within this framework of CPC, as a conceptualisation of holistic child and family-centred care that the current study is presented.

## 1.4 Classification of childhood life-limiting conditions

The identification of children who would benefit from palliative care service provision remains rather vague (Fraser et al., 2014). The United Kingdom (UK) charity Together for Short Lives has, however specified four primary categories of childhood life-limiting conditions for which palliative care may be appropriate and beneficial. In summary, the classifications include: possibly curable (Category 1), conditions associated with periods of good health, but limited-life; (Category 2), progressive conditions without curative treatment options (Category 3) and non-progressive conditions causing severe disability (Category 4). These categories are outlined in greater depth in Table 1 below. There are over three hundred conditions which fall into the life-limiting and life-threatening category (Together for Short Lives, 2013; Together for Short Lives, 2018). This results in a heterogeneous group encompassing a wide spectrum of conditions. Categorisation is not easy and the examples given are not exclusive. However the four categories have raised awareness and offered some guidance globally of the types of diagnoses where children would benefit from a palliative care approach (Bradford et al., 2012).

**Table 1: Categories of life-limiting conditions and life-threatening conditions (Together for Short Lives 2018, p.11).**

<p><b>Category 1</b> <b>Life-threatening conditions for which curative treatment may be feasible but can fail.</b> Access to palliative care services may be necessary when treatment fails or during an acute crisis, irrespective of the duration or threat to life. On reaching long-term remission or following successful curative treatment there is no longer a need for palliative care services. Examples: Cancer, irreversible organ failures of heart, liver, kidney.</p>
<p><b>Category 2</b> <b>Conditions where premature death is inevitable.</b> There may be long periods of intensive treatment aimed at prolonging life and allowing participation in normal activities. Examples: Cystic fibrosis, Duchenne muscular dystrophy.</p>
<p><b>Category 3</b> <b>Progressive conditions without curative treatment options.</b> Treatment is exclusively palliative and may commonly extend over many years. Examples: Batten's disease, Mucopolysaccharidoses.</p>
<p><b>Category 4</b> <b>Irreversible but non-progressive conditions causing severe disability, leading to susceptibility to health complications and likelihood of premature death.</b> Examples: Severe cerebral palsy, multiple disabilities, such as following brain or spinal cord injuries.</p>

However, these categories have been described as too restrictive and some suggest they need to be supplemented by identifying precise diagnoses (Hain et al., 2013; Wood et al., 2010). Children receiving care for serious injuries caused by accidents such as road traffic accidents, for example, do not fit into this method of categorisation (Pearson, Ward-Platt and Kelly, 2011). Palliative care services could, however, have a valuable role in supporting end-of-life discussions in intensive care for children with severe injuries leading to death (Hain et al., 2013). Additionally palliative care teams could provide ongoing emotional support after the death of a child (Noyes et al., 2013). In addition neonates with limited life expectancy do not fit into this method of categorisation (Wood et al., 2010). Furthermore, this method of categorisation provides a measure of objectivity but still relies on a certain degree of subjective judgement (Hain et al., 2013). For example, severe cerebral palsy or multiple disabilities may be open to different interpretations amongst healthcare professionals. This indicates, perhaps a potential value in extending the categories to reflect the broader role that might be played by palliative care services (Noyes et al., 2013). Whilst this debate may continue, the important matter is not the category, but whether all children who need palliative care, and their families, can access it regardless of their condition.

Children from all four categories of childhood life-limiting conditions will be included in this study. However, children who have a diagnosis of cancer will be excluded as both the illness trajectory and CPC services differ significantly for these children (Bergstrasser, 2018; Price, 2012). Children with cancer frequently experience a more sudden decline than children with NMLLCs who are more likely to experience slow deterioration thus the service requirement is different for these children (Nageswaran, Hurst and Radulovic, 2018; Wood et al., 2010). Children with NMLLCs are included as they account for the majority of those in need of palliative care (ACT, 2009; McCulloch, Comac and Craig, 2008; Siden, 2018). This is partly due to technological advances, resulting in growing numbers of these children experiencing improved survival (Amin et al., 2017; Wallis et al., 2011). In fact, some of these children remain dependent on the technology that has enabled them to survive for the duration of their lives. Unfortunately, many of these children will not survive to adulthood, necessitating a palliative care approach to meet their care needs throughout their lives.

## **1.5 Prevalence of children with life-limiting conditions**

It is difficult to establish the prevalence rates of children with life-limiting conditions for a number of reasons, but most importantly the diverse nature of clinical providers, and lack of agreement on important definitions (Noyes et al., 2013). Prevalence data are nevertheless, imperative in order to inform service development for children with life-limiting conditions (Fraser et al., 2014; Noyes et al., 2013). However to date, no country has a database identifying children with life-limiting conditions, thus information on the prevalence of children who need palliative care worldwide is limited. As a result, most documents and needs assessments rely on estimates on the prevalence of life-limiting conditions in children. In the absence of a national database on children with life-limiting conditions, data from a needs assessment published in 2005 continues to be utilised in Ireland (DoHC/IHF, 2005). In this needs assessment calculations for the number of children living with a life-limiting condition in Ireland were extrapolated from UK data which at the time, reported a prevalence rate of 12:10,000 child population (ACT and RCPCH, 2003). This was utilised although known to be an underestimate. Applying this ratio to Irish census data it was estimated that 1,369 children were living with a life-limiting condition in Ireland (Ling et al., 2015). Cultural factors that may impact on the accuracy of these estimates, for example, the legislation governing termination of pregnancy in Ireland at the time were not considered despite the likelihood of impacting on the number of children born and surviving with complex care needs and conditions that may result in premature death (Boyd et al., 2008). The prevalence rate of 12:10,000 child population was also utilised in Northern Ireland for many years (Fraser et al., 2011). More recent research determined that the prevalence of life-limiting conditions in children in England was double previous estimates (Norman and Fraser, 2014). Fraser et al. (2012) determined that the prevalence of life-limiting conditions in children and young people in England, at 32 per 10,000. Prevalence is highest for children aged less than one year but time trends show no increase for the youngest age group but significant increase for older children (Norman and Fraser, 2014). These increases are mirrored by a decrease in all-cause mortality for children suggesting improved survival. Rates are highest in more deprived areas and those typified by multicultural populations (Fraser et al., 2012). Norman and Fraser (2014) suggest that increasing prevalence and number of cases over time may require increased CPC service provision in the future.

## **1.6 Policy and service contexts**

The study was undertaken in and informed by policy/service contexts. The subsequent section outlines the policy and service milieu in which palliative care is currently provided to children with NMLLCs and their families in Ireland (both north and south). It is impossible to ignore the broader

international debates and perspectives concerning palliative care for children, which have impacted on the development of these policy and service initiatives. Consequently, discussion with regard to this will take place first.

### **1.6.1 International perspective**

Recent debate on the provision of palliative care as an international human right has included the assertion that there should be equity of access to services for all (Downing et al., 2016; Gwyther, Brennan and Harding, 2009). Few would argue with the merits of such an aim. Few also would argue that current evidence points to serious inequalities; at a global level, the lack of palliative care provision in many settings remains a fundamental issue (Burt, 2012). The provision of palliative care to children worldwide is currently limited to high resource countries (WHO, 2018). In 2011, a systematic review of CPC services worldwide found that 65% of countries had no known CPC. However 18% had capacity building activities, 9% were well able to cite example of local provision but only 5% had existing CPC (Knapp et al., 2011). Therefore, the provision of high quality palliative care for children and young people remains a global concern (Downing et al., 2016; Downing, Marston and Boucher, 2010). It has been well documented that there are many challenges to the provision of palliative care to children including a lack of education for families and providers; limited resources; lack of evidence; limited access to medications- including opioids for pain management and the lack of appropriate policies to ensure availability and access within the healthcare system (Downing et al., 2018). According to Marston and Chambers (2012) even in rich resource countries, the lack of sustainable funding and problems with coordination of services means that palliative care for children remains patchy and inequitable. Additionally there is evidence that some groups are excluded from services, or have special needs that are not met. These can include the less affluent and children (WHO, 2018). The reasons for variations in access are unclear.

### **1.6.2 Children's palliative care in the United Kingdom**

While many countries are in the process of developing CPC, the UK has led the way in developing CPC as an internationally recognised specialty (Drake, 2018; Hain, Heckford and McCulloch, 2012). Palliative care services specifically for children developed initially from children's oncology services and more recently as palliative care teams in their own right. Together for Short Lives (previously the Association for Children with Life-threatening or Terminal Conditions (ACT) and the Royal College of Paediatrics and Child Health (RCPCH) in the UK has been instrumental in producing several key documents regarding the palliative care needs of children with life-limiting conditions. An

independent review of the long-term sustainability of CPC services was undertaken by the Department of Health in 2006 (Craft and Killen, 2007). This review highlighted that CPC services in England developed over the past twenty-five years in a largely unplanned and ad hoc way, often as a result of both local interest and the availability of charitable funds. This has resulted in significant variation in the structure, location and format of services across England (Craft and Killen, 2007). As a result of this independent review a national strategy on CPC was developed (DoH, 2008). Standards for CPC services have also been developed in Scotland and Wales (Noyes et al., 2013). These documents informed the development of CPC policy in Ireland (DoHC, 2009).

### **1.6.3 Background to Palliative Care Policy and service in Ireland**

Palliative care services for children in Ireland have been the focus of considerable change in recent years, partly in response to increased recognition of the needs of children with life-limiting conditions and their families and the fact that commitment to improving care and future services has been placed high on the political agenda (DoHC, 2009). The *Report of the National Advisory Committee on Palliative Care* (DoHC, 2001) laid out a blueprint for palliative care development in Ireland (Culliton, 2009). Since then, this report has been adopted as a national policy (DoHC, 2009). Although primarily focused on adult palliative care, several recommendations were made regarding CPC. The report identified the need to maintain the best quality of life possible, incorporate palliative care as early as possible and emphasised that palliative care should be available to every child and family. This report made several recommendations regarding the delivery of palliative care services based upon the principles of equity, accessibility and the adoption of a coherent and comprehensive national policy framework for palliative care. Recommendations included that palliative care is best provided in the home and palliative care services for children, including respite care, should be provided as close to the home as possible.

However, the *Children's Palliative Care Needs Assessment* (DoHC, 2005) identified several ongoing problems in relation to CPC services in Ireland. These included issues of access; access to palliative care services in Ireland was dependent on the child's geographic location and diagnosis. It also highlighted the lack of CPC services within the health system, with some children receiving care from adult specialist palliative care services. Numerous other problems were outlined, including fragmentation of services, lack of service coordination and a lack of education and training for healthcare professionals caring for children with life-limiting conditions. Furthermore, there were no data on the number of children living with and dying from life-limiting conditions in Ireland. The report highlighted the need for a nationwide CPC service to ensure equity in service provision,

greater coordination of services, development of home care and community services and accurate data collection regarding prevalence of life-limiting conditions and mortality. In recognition of the inadequacies in the system, the DoHC (2009) report *Palliative Care for Children with Life-Limiting Conditions in Ireland –A National Policy* detailed how palliative care services should be developed for children with life-limiting conditions and their families, and emphasised that all children should have the choice to be cared for at home with the support of a multidisciplinary team. This policy is aimed at addressing the issues identified in the needs assessment in order to construct a CPC service that is responsive to the needs of children and their families. It clearly states that palliative care services for children should be accessible, equitable, flexible and appropriate, and should meet the needs of every child with a life-limiting condition and their family. However the timeframe for implementation is not outlined in the policy document.

The National Clinical Programme for Paediatrics and Neonatology has developed a *Model of Care for Paediatric Healthcare Services* in Ireland. The model sets out to underpin the delivery of healthcare for children both in the present and into the future. Key steps in its development were wide consultation with all healthcare professionals, involvement with parents and parent groups, a detailed analysis of the current clinical activity of all paediatric medical and surgical subspecialties, the current interface between primary, secondary and tertiary care services, and a study of how paediatric clinical care is distributed with reference to international best standards. The policy was<sup>1</sup> developed from the ground up and involvement and advice was sought from those working in hospital management and community services throughout the process. Paediatric Palliative Care is specifically addressed within this model of care. It is suggested that current paediatric palliative care services are insufficient to meet increased needs. In order to provide a quality, equitable service, development will be required over a number of phases. Short, medium and long term goals are revealed within this model of care. Please see link below.

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<sup>1</sup> **PDF Link available online**

<https://www.hse.ie/eng/services/publications/clinical-strategy-and-programmes/paediatric-palliative-care.pdf>

#### **1.6.4 Northern Ireland policy and service context**

A number of strategic policies and guidelines have directly and indirectly contributed to the development of palliative care and end-of-life care services for children in Northern Ireland. An *Integrated Care Pathway for Children with Complex Physical Healthcare Needs* provided guidance in meeting the needs of children and young people who have complex physical health and social care needs (DHSSPS, 2009). It also recognises the needs of their families. A review of CPC services and end-of-life care took place in Northern Ireland. It is the third phase of a wider Department of Health, Social Services and Public Safety Review of Health and Social Care (HSC) paediatric services which aims to provide a strategic direction for the future development of HSC services for children and young people over the next ten years (DHSSPS, 2014). The Children's Palliative Care Strategy - *A Strategy for Children's Palliative and End-of-Life Care 2016-2026* was launched in November 2016 (DoH, 2016). This document sets out the strategic direction for the palliative and end-of-life care of ill and dying children and young people in the North for the next ten years. It is suggested that the success of this Strategy can really only be measured through outcomes; the real-life experiences of the children, families and carers who live on a daily basis with the reality of a child that needs palliative or end-of-life care. Therefore, the Strategy adopts an outcomes-based approach to palliative and end-of-life care for children. The aim is to provide quality, safe, sustainable and appropriate palliative and end-of-life care to ensure the best possible outcome for the child and their family. While the focus of the Strategy is on the child, the objectives also seek to help and improve the experience of the parents, carers, and wider family circle. Primary, secondary, tertiary, community and voluntary sector care professionals are all partners in this care. The unique role of the family must be recognised in the ongoing care. Decisions are made in conjunction with the parents and the child, and are in the child's best interests. Progress on the implementation will be monitored through new transformation accountability arrangements. However, it is also acknowledged within the Strategy that the objectives will also be subject to available resources against the background of competing priorities for new investment across health and social care. A *Strategy for Paediatric Healthcare Services Provided in Hospitals and in the Community 2016-2026* has also recently been published, focusing on acute hospital services, the management of transition of such services into adult services, and the interface between hospital and community services (DoH, 2016).

#### **1.6.5 Policy and service context Ireland and Northern Ireland**

Palliative care services for children in Ireland and Northern Ireland have been the focus of considerable change in recent years. This is partly in response to increased recognition of the needs

of children with life-limiting conditions and their families and the fact that commitment to improving care and future services has been placed high on the political agenda in both jurisdictions (DOH, 2016; Jordan and Fullerton, 2016). A number of strategic policies and guidelines have directly and indirectly contributed to the development of palliative care and end-of-life care services for children in Ireland and Northern Ireland (DoHC, 2009; DHSSPS, 2014; DoH, 2016). Similarities exist between the policies and guidelines in both jurisdictions. It is suggested that the success of the current strategies can really only be measured through outcomes; the real-life experience of parents and families who live on a daily basis with the reality of a child that needs palliative care or end-of-life care. While the focus is on the child, the objectives also seek to help and improve the experience of the parents and families. Improved co-ordination of services for children with life-limiting conditions are emphasised. Additionally, it is suggested that parents should be actively involved in decision-making and planning for their child. There are however some notable differences in CPC service provision between Ireland and Northern Ireland. For example, the greater level and availability of hospice services for children with life-limiting conditions in Northern Ireland than for children in Ireland (Price, McCloskey and Brazil, 2017; Quinn and Hillis, 2015).

## **1.7 Thesis conspectus**

This thesis consists of nine chapters, outlined as follows. Chapter two presents a review of the existing literature on palliative care for children in particular children with NMLLCs. The theoretical framework on which this study is based is presented within this chapter. Chapter three presents the study critiques. The rationale for the present study and the aims and objectives are stated at the end of this chapter. Chapter four presents the methodological approach adopted, namely a mixed methods design. Chapter five presents the qualitative and quantitative research findings from the parents in this study. In Chapter six the qualitative and quantitative research findings from the parents are critically discussed with reference to the initial study aims and objectives and set in the context of the current evidence base. In Chapter seven the qualitative research findings from the healthcare professionals are presented. In Chapter eight the qualitative research findings from the healthcare professionals are critically discussed with reference to the initial study aims and objectives and set in the context of the current evidence base. To conclude, Chapter nine presents an integration of the qualitative and quantitative research findings from the parents and the qualitative research findings from the healthcare professionals. The original contribution the present study makes will be considered. This final Chapter also presents the strengths and limitations of the study and implications for practice, future research, education and policy.

## **Chapter 2: Literature Review/Theoretical framework**

### **2.0 Introduction**

This chapter presents a review of the research literature related to the subject area of the study. The literature reviewed encompasses a wide range of sources, including empirical research employing a diverse range of methodologies and is befitting with what has been identified as an integrative literature review (Lo-Biondo-Wood and Haber, 2017; Sparbel and Anderson, 2000; Toracco, 2005). Maintaining scientific integrity while conducting an integrative literature review involves careful consideration to threats to validity (Russell, 2005). De Souza, Silva and Carvalho (2010) developed an explicit and systematic approach to integrative literature reviews comprising of 6 stages: formulate the guiding question, search literature and include, extract data from studies, analyse studies critically, discuss results and disseminate the study findings. The literature review, which follows, is underpinned by the principles of the De Souza, Silva and Carvalho (2010) integrative framework.

### **2.1 Defining an integrative literature review**

An integrative literature review is conducted to appraise the quality of research, discover gaps in what is known, infer generalisation of a phenomenon, identify central themes and make connections between related areas of specialisation (Christmals and Gross, 2017). The integrative review method is the only approach that allows for the combination of diverse methodologies (De Souza, Silva and Carvalho, 2010). It helps to formulate research questions, identify theoretical frameworks in a research area, propose the need for future research and explore the research methodology used (Cooper, 1982; De Souza, Silva and Carvalho, 2010; Ganong, 1987; Russell, 2005, Torraco, 2005; Whitemore and Knafl, 2005). It is suggested that all integrative review frameworks make tremendous contributions to integrative review methodology (Cooper, 1982; De Souza, Silva and Carvalho, 2010; Ganong, 1987; Russell, 2005; Torraco, 2005; Whitemore and Knafl, 2005). Yet, all of the frameworks lacked clarity with regard to structuring the integrative review in particular the positioning of the theoretical framework within the review. The theoretical framework on which this study is based is presented towards the beginning of this integrated literature review.

### 2.1.1 Review aim

The aim of the following review was to systematically examine the evidence surrounding the experience of caring for children with NMLLCs including, the experience of accessing services and decision-making surrounding the child's care.

The specific questions of the review were:

1. What are the experiences of parents caring for a child with a NMLLC including the experience of service provision?
2. What are parents' views of accessing services when caring for their child?
3. What are the experiences of parents and healthcare professionals with regard to decision-making?
4. What are the experiences of healthcare professionals caring for children with NMLLCs including their experience of service provision?

## 2.2 Search strategy

Once the question for review has been formulated, De Souza, Silva and Carvalho (2010) suggest that the literature search can begin. 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. The last search was carried out in June 2019. Electronic databases included: Academic Search Complete, Cumulative Index of Nursing and Allied Health Literature (CINAHL), MEDLINE, PsycArticles and PsycINFO. The database Dissertation Abstracts was used to identify unpublished theses in the area. Literature from voluntary organisations, including Together for Short Lives and the Irish Hospice Foundation (IHF) were reviewed as were official relevant Government publications for both Republic of Ireland and Northern Ireland. Search engines such as Google Scholar and PaedPal Lit were also used. No date parameters were applied to the literature search at the outset. The search was confined to English language material only. In association with the term '*palliative care*' and '*children*', the initial keywords *life-limiting* and *life-threatening* were used. In the context of the difficulties associated with terminology other key related terms such as *medically fragile* and *rare conditions* were also used in association with key words. Boolean operators 'or' and 'and' were used to connect and define the relationship between the key terms and keywords. While the search strategy incorporated a broad view of the literature to begin with ultimately this became more refined to include experiences, access and decision-making. This involved manually sifting through the literature. De Souza, Silva and Carvalho (2010) propose that the search strategy should be broad and diverse to begin with. Precise details regarding the search strategy used are demonstrated in Table 2.

**Table 2: Terms and keywords utilised in the search strategy**

Key terms	Key terms	Keywords
<b>Boolean operators</b>	<b>Boolean operators</b>	<b>Boolean operators</b>
<b>'or'</b>	<b>'or'</b>	<b>'or'</b>
Palliative care	Children	<i>Life-limiting</i>
End-of-life care	Child	<i>Life-threatening</i>
End of life care	Infant	<i>Life-shortening</i>
Terminal care	Neonate	<i>Medically fragile</i>
Hospice care	0-18 year olds	<i>Rare conditions</i>
Technology	0-18 y/o	<i>Duchenne muscular</i>
dependent	Adolescent	<i>dystrophy</i>
Complex care		<i>Cerebral palsy</i>
		<i>Batten's disease</i>
		<i>Mucopolysaccharidoses</i>
<b>Boolean operators</b>		
<b>'and'</b>		

### 2.2.1 Search results

A large number of hits were found in the bibliographic databases. The results are revealed in Table 3.

**Table 3: Hits from online bibliographic databases**

Bibliographic databases	Number of hits
Academic Search Complete	473
CINAHL	689
MEDLINE	754
PsycArticles	2
PsycINFO	394

Much of the early literature presented commentaries on care management for children who are medically fragile/technology dependent and on expanding roles and responsibilities in the

community as opposed to explicit research based papers. Though, from 1990 onwards the publication of research based papers became more prominent. However, the majority of these papers focused on children with cancer as opposed to children with NMLLCs. From 2000 onwards the literature began to incorporate children with NMLLCs; however the predominant attention remained on children with malignant conditions. Even in a recent analysis of studies concerning pediatric palliative care Flexa Souza et al. (2018) showed a literature focused on oncology and caregiver care, revealing still low publication of other chronic conditions in pediatrics. Studies focusing exclusively on children with cancer were excluded from the literature review as both the care trajectory and CPC services differ significantly for these children. Additionally, studies focusing on children with disabilities as distinct from children with life-limiting and life-threatening conditions were likewise excluded for the same reason. Whilst some elements of their care overlap, other elements have nuanced differences. Studies concerning complementary and alternative therapies were also excluded as these did not relate specifically to care issues for children with NMLLCs and their families. Papers reporting on issues peripherally associated with the topic area for example, palliative care economics and impact of research participation were not included in the literature review. As part of what De Souza, Silva and Carvalho (2010) describe as extract data from studies, a total of one hundred and twenty seven studies met the criteria and were included in this review. This incorporated studies with reference to the experience of caring for children with NMLLCs including, the experience of accessing services and decision-making surrounding the child's care.

Details were extracted from each study, to include: author(s), originating country (based on first author affiliation), aim/objective, study design, sample, data collection techniques and key findings (Appendix N). De Souza, Silva and Carvalho (2010) suggest that this approach, in addition to enabling a brief organisation of data, facilitates comparison among studies in specific topics, such as problems, variables and characteristics of the sample. The studies have been colour coded with reference to experiences, access and decision-making. Frequently issues surrounding experiences, access and decision-making spanned across individual studies. However, the predominant focus incorporated one topic area and therefore these were colour coded accordingly. The studies primarily focusing on parents/families are presented first in the study summaries. Subsequently the studies focusing on healthcare professionals are presented. Finally, studies which include both parents/families and healthcare professionals are presented.

**Table 4: Coding of studies with reference to experiences, access and decision-making parents/families**

Experiences	Access	Decision-Making
41	13	14

**Table 5: Coding of studies with reference to experiences, access and decision-making healthcare professionals**

Experiences	Access	Decision-Making
15	5	6

**Table 6: Coding of studies with reference to experiences, access and decision-making parents/families and healthcare professionals**

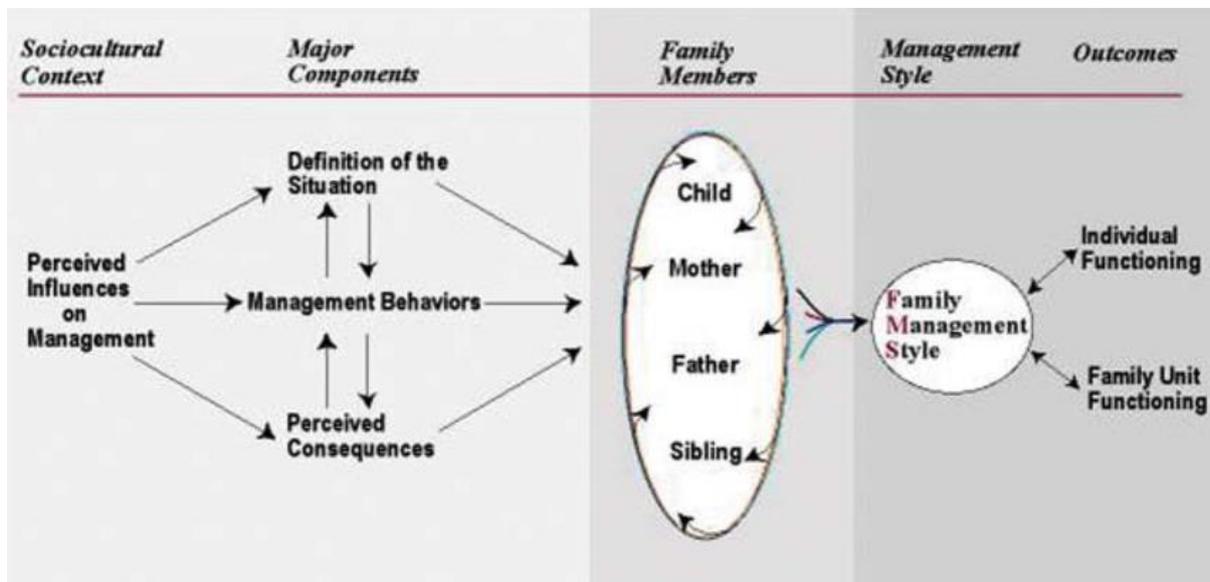
Experiences	Access	Decision-Making
8	13	12

### 2.3 Theoretical framework

A theoretical base was needed to underpin this study, which reflected the critical concepts with which the study was concerned. This study is entrenched in the concept of CPC which accentuate the child and family as a unit of care, the delivery of family-centred care and a focus on the family. In seeking to source an appropriate theoretical framework numerous theories, concepts and models were considered and reviewed in the areas of nursing and psychology. The Family Management Style Framework (FMSF) developed by Knafl and Deatrick (2003) and Paterson’s (2001) Shifting Perspectives Model of Chronic Illness were reviewed and chosen. The FMSF (please see Figure 1) provides a description of how families manage both family life and their children’s complex health problems (Deatrick et al., 2006). Families, to varying degrees, strive toward the incorporation of normalisation, depending upon each family’s definition of the situation, in the way they manage their children’s complex care needs, and perceived future consequences of a child’s condition-both for the child and family life (Deatrick et al., 2006). Most families who have children with serious illnesses eventually view their children and their lives as normal and manage the illness-related demands successfully (Deatrick et al., 2006). However, not all families see their lives as normal, and

those who do reach this point often use a variety of strategies over time as they undergo a continual process of adjustment. Normalisation efforts are thought to affect outcomes such as individual and family functioning (Knafl and Deatrick, 2006). The FMSF is narrower in scope than frameworks addressing overall family functioning or family coping and adaptation to a stressor. However, the FMSF is broader in scope than conceptualisations of family response addressing a specific condition or a single aspect of family response such as decision-making (Knafl, Deatrick and Havill, 2012). The FMSF is recognised as a useful framework for furthering knowledge of family response to childhood chronic illness (Van Riper, 2010). Bousso et al. (2012) utilised the FMSF to explore how families define and manage their lives when they have a child or adolescent undergoing palliative care at home. This research supports the usefulness of the FMSF in pediatric palliative care. Using the FMSF to understand how families manage can provide healthcare professionals with insights on how to work effectively with families (Bousso et al., 2012). For example, the FMSF can be used to identify key aspects of families' efforts to incorporate illness management into everyday life, and to identify strengths and care challenges to target for intervention. Without such knowledge, their efforts to guide and support families throughout the care trajectory through the appropriate provision of services remain limited.

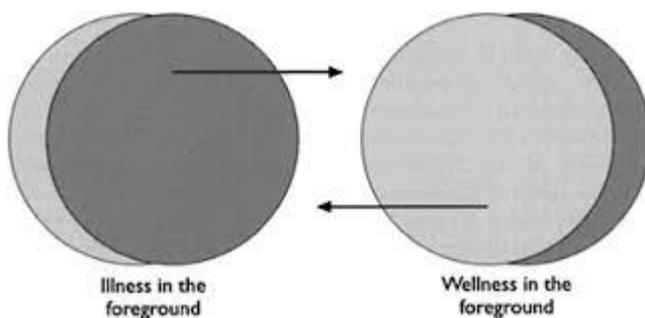
**Figure 1: Family Management Style Framework**



Paterson's (2001) Shifting Perspectives Model of Chronic Illness (please see Figure 2) proposes that living with a chronic illness is an ongoing, continually shifting process whereby the illness is sometimes in the foreground, and at other times, in the background. When illness is in the

foreground, the focus is on the illness and the suffering and loss that come with the illness such as the negative effect on family members. But when it's in the background the family focus shifts to the health and well-being of the entire family. Accordingly Paterson's (2001) Shifting Perspectives Model of Chronic Illness highlights the importance of perception. Paterson's Shifting Perspectives Model of Chronic Illness was previously used to illuminate the meaning of living as a family in the midst of chronic illness (Årestedt, Persson and Benzein, 2014). It was also used to examine family functioning and normalisation in mothers of children dependent on medical technology (mechanical ventilation, intravenous nutrition/medication, respiratory/nutritional support) following initiation of home care (Toly, Musil and Carol, 2012). Ongoing illness adaptation influences individuals' decisions and in particular the decision to utilise healthcare services (Williamson, Koro-Ljungberg and Bussing, 2009). Thus Paterson's Shifting Perspectives Model of Chronic Illness is fitting to help understand how families approach decision-making and in particular the decision to access and utilise healthcare services. The FMSF and Paterson's Shifting Perspectives Model of Chronic Illness recognise 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.

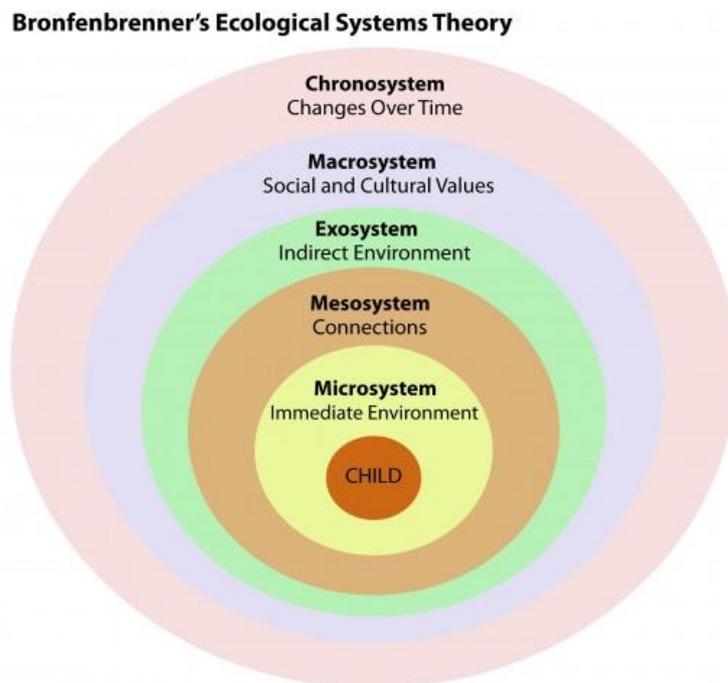
**Figure 2: Paterson's (2001) Shifting Perspectives Model of Chronic Illness**



However, it is also critical to establish the context families live in, in order to truly understand their experiences. An ecological perspective offers a way to simultaneously emphasise both individual and contextual systems and the interdependent relations between these two systems. Thus the Ecological Systems Theory developed by Bronfenbrenner (1979) was also selected to guide this study. In the ecological model, the complex interplay between the child or young person, the family and their social and physical environment is represented in a number of systems. These are diagrammatically represented in Figure 3 and described beneath. The potential of this theory to the current study lay in its emphasis on studying relationships amongst family subsystems from the

Microsystem of the immediate family through to socio-political and economics in the Macrolevel. Furthermore, palliative care services for children exist within an ecological context; therefore, it is essential to adopt an ecological viewpoint. Importantly the child is at the centre of this model which is congruent with the child-centred perspective. The model acknowledges that a child affects as well is affected by the setting in which he/she spends time. Chandran, Corbin and Shillam (2016) used Bronfenbrenner's Ecological Systems Theory to explore caregiver experiences in adult palliative care.

**Figure 3: Bronfenbrenner's Ecological Systems Theory**



### **Microsystem**

This is the layer closest to the child and contains the structures with which the child has direct contact. The microsystem embodies the child's immediate physical environment and the relationship between family members and other individuals that the child is in direct contact with. At this level, relationships have impact in two directions-both away from the child and toward the child. This is the most influential layer of the ecological system theory.

### **Mesosystem**

The mesosystem links and makes connections between microsystems directly involving the child for example, home, school or the social care setting. For children with NMLLCs this may include the hospital.

### **Exosystem**

This layer defines the larger social system in which the child does not function directly. The structures in this layer impact the child's development by interacting with some structure in her microsystem (Berk, 2009). Parent workplace schedules or community-based family resources are examples. The child may not be directly involved at this level, but he does feel the positive or negative force with the interaction with his own system.

### **Macrosytem**

This layer may be considered the outermost layer in the child's environment. The effects of larger principles defined by the macrosystem have a cascading influence throughout the interactions of all other layers. For example, if it is the belief of the culture that parents should be solely responsible for raising their children, that culture is less likely to provide resources to help parents. This, in turn, affects the structures in which the parents function. The parents' ability or inability to carry out that responsibility toward their child within the context of the child's microsystem is likewise affected. Palliative care policy is significant in the Macrosystem.

### **Chronosystem**

This system compasses the dimension of time as it relates to a child's environment.

Resilience is not a dimension that is included in the Bronfenbrenner (1979) model. It has been argued that it should be integrated into the model as it gives a better understanding of an individual's capacity for overcoming negative influences and situations (Engler, 2007). It is important to know how the family defines the situation and manages family life in different contexts. The FMSF and Paterson's Shifting Perspectives Model of Chronic Illness are valuable in describing contextual factors of the experiences of families. Contextual factors are important in providing healthcare professionals with insights on how to work effectively with families. For example, it is important that services are identified and understood to meet the needs of these children and families. Consequently the amalgamation of the FMSF, Paterson's Shifting Perspectives Model of Chronic Illness and Bronfenbrenner's Ecological Theory were considered appropriate. These informed decisions made in the research process. For instance, the decision to include both parents and healthcare professionals in the study and decisions regarding the topics and questions included in the interview guides.

## **2.4 Concluding comments**

This chapter has presented a review of the research literature related to the subject area of the study. The search strategy is clearly identified. The theoretical framework on which this study is based is presented within this chapter. The study critiques will be presented in the subsequent chapter. Additionally the rationale for the present study and the aims and objectives will be outlined at the end of the said chapter.

## **Chapter 3: Literature Review/Study critiques**

### **3.0 Introduction to study critiques**

In this section the studies which met the inclusion criteria of the research will be critically discussed. As previously suggested this includes studies with reference to the experience of caring for children with NMLLCs including, the experience of accessing services and decision-making surrounding the child's care. De Souza, Silva and Carvalho (2010) suggest that the studies should be divided into subgroups according to a previously established classification aiming to facilitate the analysis. To commence studies with reference to experiences will be critiqued. Subsequently studies with reference to access and decision-making will be appraised. Interim comments will be presented following each section.

### **3.1 Parents/families experiences including their experience of service provision**

Caring for a child with a NMLLC is a complex experience, which can often result in crises, followed by periods of change, readjustment and periods of relative stability but where there is uncertainty as to when the next crisis will occur (Bally et al., 2018; Carter, 2017; Menezes, 2010). Thus parental caregiving for these children can be a very involved process, with losses of significant milestones occurring throughout the care trajectory (Cadell, Kennedy and Hemsworth, 2012). Regardless of diagnosis or prognosis, parents are central to the care that children receive, both at home and in healthcare settings (Cadell, Kennedy and Hemsworth, 2012; Carter, 2017). Thus, it is imperative to understand the experiences of parents caring for a child with a NMLLC including their experience of palliative care service provision.

#### **3.1.1 Caring over the course of the trajectory**

Caring for a child with a NMLLC at home over prolonged periods can be physically and emotionally demanding on parents (Knapp et al, 2010; Monterosso et al., 2007; Price et al., 2012). Parents are expected to assume many roles that extend across physical, emotional, social, and spiritual domains, including, everyday active care provider, decision-maker, advocate, nurse, relationship manager, care coordinator, communicator, and parent (Caicedo, 2014). The need for palliative care is often prolonged due to technical and medical improvements (Verberne et al., 2017). Coad, Patel and Murray (2014) suggest that the intensity of caring places an immense burden on parents. This frequently results in parents becoming socially isolated from family and friends (Eaton, 2008; Knapp

et al., 2010). It has been reported that whilst parents may initially receive support from friends and family members, this decreases over time as increased training and responsibility is needed (Collins et al., 2016; Cote-Arsenault and Denney-Koelsch, 2011). Parents often find it difficult to find someone reliable to care for their children (Knapp et al, 2010). Furthermore, the financial impact of caring for a child with a NMLLC is significant (Beaune et al., 2014; Nicholl et al., 2013; Normand and Revill, 2010; Steele and Davies, 2006).

Rallison and Raffin-Bouchal (2013) undertook a hermeneutic inquiry using in-depth interviews to explore the experience of six families (13 interviews) living with and caring for a child with a progressive neurodegenerative illness in Canada. Family members in the study included parents, siblings, the ill child, and other significant individuals that the family identified as their family. The three main themes that emerged were the families' experiences of holding predictability and unpredictability, the complexity and relentless care in the home and the spiritual journey of the families. The families began to realise early in their child's life that the child they had hoped for was ill. Families faced a long and difficult search for a diagnosis which sometimes took months or years. Sometimes the specific diagnosis was never found. When a diagnosis was made, frequently healthcare professionals had never heard of these rare conditions and did not know what to expect. The families of these children struggled to find someone who could help guide them on their journey. Families experienced an overwhelming sense of fear in the early days of living with their ill child. All of the families spoke of exhaustion, of the relentlessness of caring for their children, which left little time for sleep. Although there were periods of stability in the child's life these were overshadowed by fear that there would be a crisis ahead. The intensity of care in the home through the lengthy trajectory of the child's illness often resulted in families becoming housebound. The findings echo those of previously mentioned studies (Caicedo, 2014; Coad, Patel and Murray, 2014; Knapp et al, 2010; Monterosso et al., 2007; Price et al., 2012). Families eventually discovered that funding was available for respite care. Trusting others to share the care of the children was challenging. The cost to the families of caring for the children was also significant. Expenses included the cost of medications, supplies, special equipment such as wheelchairs and breathing machines, and modifications to houses and/or vehicles. Families received funding from various sources, but many spoke of inconsistencies, and much of it was dependent on the family's ability to write proposals and to lobby for funding. Families in this study spoke of the importance of their relationships with care providers. However the majority of the children and families were not connected with a consistent interdisciplinary team with whom ongoing, long-term relationships

could be forged and fostered. Although this study provides important insights, the diversity of the sample limits the insights to a certain extent.

In a recently published study Courtney et al. (2018) explored mothers' perspectives of the experiences and impact on themselves and their family when their child has a life-limiting neurodevelopmental disability in Ireland. Twelve mothers were interviewed and topics included mothers' experiences of caring, the impact on themselves and their family of care provision, and the management of day-to-day life. Four themes were identified. 'Starting Out' relates to mothers' experiences of the birth of their child and the aftermath. 'Keeping the Show on the Road' describes the strategies families employ to manage life day to day and the resources they use. 'Shouldering the Burden' describes the range of physical, psychological, and social consequences of the situation for mothers and the family. 'The Bigger Picture' relates to the world outside the family and how this is navigated. Bronfenbrenner's theory reminds us how important it is to think about the 'big picture' when caring for children with NMLLCs and their families including how the environment and relationships affect them. Yet the contextual factors of the experiences of mothers are also pertinent. The findings suggest mothers' overall experiences are characterized by a constant struggle, with evidence of negative impacts on family life, though there was also evidence of resilience and coping. Mothers reported constant struggles with services. This mirrors the findings from other studies in which parents spoke about 'fighting the system' and their 'battle' for adequate service provision (Dybwik et al., 2011; Price et al, 2012; Rallison and Raffin-Bouchal 2013; Somanadhan and Larkin, 2016). Parents also described the constant worry and anxiety they experience about the future, including the worry that services would be withdrawn or reduced. The authors identified several limitations to this study. Participants were recruited through a single service provider, which may limit the generalizability of the findings. In addition, as all children represented were under six years and the majority had received a diagnosis based on a problem at birth or soon after, it is possible that the experiences of this group may not reflect those of parents who experience a later diagnosis. However, access to families can be a challenge in general, and this study is positioned to provide some insights into their experiences. The lack of gender representativeness is a second limitation, as this study presents mothers' narratives and is therefore confined to mothers' experiences.

Menezes (2010) undertook a grounded theory study to explore the experiences of families (39 participants from 10 families) affected by life-limiting conditions in the United Kingdom. Participating family members included children with life-limiting conditions, siblings and parents.

This study identified several 'moments of realization', representing times in the child's life when participants recognised the real threat to the child's life, these included: questions of inheritance, diagnosis and prognosis, acute loss of abilities, slow deterioration and life-threatening surgery. This research found that uncertainty was a recurring theme that rose and fell within the family unit and across individual family members. This finding was likewise reported in a previously mentioned study undertaken by Rallison and Raffin-Bouchal (2013). Sometimes there were long periods of stability in the child's health and at other times, crises came quickly or repeatedly. Parents in this study called for a better system of support for the whole family. The author concluded that it is impossible to describe the pain and isolation of families caring for children with life-limiting conditions where in the end families' face the loss of their child. Daily hurdles were innumerable and the researcher witnessed more hurdles than any family articulated. Families were recruited through a children's hospice and care team members within the hospice recruited them. The author acknowledged that it was difficult to assess the extent to which families might have felt obliged to participate in the study.

Collins et al. (2016) explored the lived experiences of parents who are currently providing care for a child with a life-limiting condition in Australia. Semi-structured interviews were conducted with 14 parents (12 mothers and 2 fathers) who identified as the 'primary caregiver' for one or more children diagnosed with a life-limiting condition. Four key themes represented the prevalent experiences of parents: (1) trapped inside the house, (2) the protector, (3) living with the shadow and (4) travelling a different pathway. They describe parents' physical and social isolation, exclusion from the workforce, pervasive grief and associated impacts to their health and well-being. Limited professional and diminished social supports resulted in full ownership of care responsibility. The findings suggest a mismatch between services offered, current standards of care and recognition of caregivers' difficulty or sometimes inability to utilise supports outside the home. The authors acknowledge that while a full commentary on possible service improvements is outside the scope of this study, the experiences shared by caregivers point to some important service considerations. This study has several limitations. As a descriptive qualitative study, the sample size was small. In addition, despite being open to both parents, the sample included mostly mothers, given very few fathers' self-identified as the primary caregiver.

### **3.1.2 Measuring the caregiving demands**

A recent Italian study of thirty-three families who cared for children with life-limiting conditions showed parents spent an average of nine hours a day meeting the child's care needs (Lazzarin et al.,

2018). This was without taking into account the other normal activities that the parents engaged in with their children, such as playing, entertainment and schooling. The children needed an average of five different life-supporting medical appliances. The time taken to provide their care increased significantly with each additional appliance. Their most time-consuming daily needs were feeding and support when they woke at night. The families' experiences were investigated using an *ad hoc* structured interview with questions that concerned the procedures and medical appliances used at home and the amount of time, in minutes, spent on daily care. The families investigated in the present study were under the care of a specialist palliative care centre that operates in cooperation with a network of local healthcare services. The expertise of the various professionals involved enabled healthcare to be provided according to a regular schedule, but daily support for the families was only guaranteed while their child was attending school or if specific projects were developed. Very often, families had to make their own arrangements, so that one parent could continue to work while the other, usually the mother, gave up her job to care for the child. This choice often caused the family additional financial problems. This reflects the findings from other studies which suggest that the financial impact of caring for a child with a life-limiting condition is significant (Beaune et al., 2014; Nicholl et al., 2013; Normand and Revill, 2010; Rallison and Raffin-Bouchal, 2013; Steele and Davies, 2006). This is compounded by a lack of services. The children investigated in the study had conditions that could be classified into the four primary categories of childhood life-limiting conditions (Together for Short Lives, 2018).

### **3.1.3 Experiences of fathers**

In terms of roles, research suggests that the role of primary caregiver is often assumed by mothers (Smith, Cheater and Bekker, 2015). Hence, research is regularly dominated by the perspectives of mothers (Courtney et al., 2018; Goldstein et al., 2013; Jennings and Nicholl, 2014). However fathers' involvement in and influence on the health and development of their children have increased in a myriad of ways in the past ten years (Yogman and Garfield, 2016). A small number of studies have been conducted to reflect this. Bailey-Pearce et al. (2017) explored fathers' experiences of their child's life-limiting illness in the United Kingdom. In addition, fathers' attachment strategies were assessed using the Adult Attachment Interview. A narrative approach was adopted. Seven fathers of children diagnosed with a life-limiting condition for at least one year participated. The dominant themes were found to be 'experience of the diagnosis', 'living with the illness', 'struggling with emotions' and 'relationship with staff'. Within each theme, there were differences which related to the father's attachment strategies. This was particularly evident in parts of their narratives recounting critical moments of threat and anxiety in the course of discovering and adjusting to their

child's illness. Importantly, the findings also suggested that the experience for the fathers stressed, and in some cases disrupted, their attachment coping strategies. All fathers told stories of trying to get it right for their children and family. Their experiences of, and adjustment to, the illness were related to their attachment strategies. All fathers talked about the bonds they had formed with staff. Broadly, fathers talked about positive experiences with staff; however fathers also described frustration with some interactions and particular professionals. Many of the fathers suggested that they were positioned on the periphery in comparison with the mothers. Some drew on culturally gendered ideas and suggested the presence of such an issue was outdated.

Nicholas et al. (2016) likewise explored fathers' experiences of their child's life-limiting illness in Canada. Additionally, their perceptions regarding support needs were explored. In-depth semi-structured interviews were conducted with eighteen fathers of children with life-limiting illness (6 fathers had experienced the death of their child). The overarching themes were stresses, means of coping, and perceived needs for support. Generally, fathers struggled relative to discursive and internalised notions of fathers as providers and protectors for their children, combined with an inability to ease their child's vulnerability to life-limiting illness. Fathers were engaged in the care of their child with life-limiting illness, but several felt marginalised by healthcare providers in care planning and staff/family communication. Some fathers felt that the system did not offer sufficient support to their child, family, and/or self, relative to the demands and strains imposed by the child's condition. Fathers described end-of-life and health care policies that impeded support access, such as age requirements and other limiting criteria that restricted assistance as needed. This study provides an invaluable insight into father's perspectives of living with and losing a child to a life-limiting illness and their perceptions regarding support needs. The number of fathers who participated was significant. The primary diagnoses of the children varied—including cancer, cystic fibrosis, cerebral palsy, enzyme deficiency, and congenital heart defects.

### **3.1.4 Interim comments in relation to parents/families experiences including their experience of service provision**

From the literature, it is apparent that caring for a child with a NMLLC is a multi-faceted experience for parents and families. Although the studies reviewed are enlightening they were largely conducted outside of Ireland, where services and care provision differ for these children (Bailey-Pearce et al., 2017; Collins et al., 2016; Lazzarin et al., 2018; Menezes, 2010; Nicholas et al., 2016). In addition, many of the studies focused on the complexities of caring with little emphasis on service provision (Bailey-Pearce et al., 2017; Menezes, 2010). Consequently the relevance of the findings

may be somewhat limited. Furthermore the research is dominated by the perspectives of mothers (Courtney et al., 2018; Goldstein et al., 2013; Jennings and Nicholl, 2014). It is also important to explore fathers' experiences. Palliative care services for children in Ireland are rapidly developing (DoHC, 2009; DoH, 2016; HSE, 2018). As CPC services develop, it is important to ascertain if the services are meeting the needs of children and parents. Thus, it is important to explore the experiences of Irish parents (both mothers and fathers) who are currently using CPC services and ascertain their views regarding service provision.

### **3.2 The experiences of healthcare professionals caring for children with NMLLCs including their experience of service provision**

It is similarly essential to explore the experiences of healthcare professionals who are caring for children with NMLLCs in order to provide an in-depth understanding of their experiences including their experience of service provision. This will allow for a more comprehensive picture of current palliative care practice. The following section will review and critique the key research that has been carried out with healthcare professionals and the major themes identified from the literature.

Bergstraesser et al. (2013) undertook a study in Switzerland in an effort to describe the perceptions and needs of pediatric healthcare professionals taking care of children with palliative care needs and to develop a concept for the first Center of Competence for Pediatric Palliative Care. Semi-structured interviews were conducted with seventy-six healthcare professionals, including physicians, nurses, psychologists and social workers. The main interview topics were: (1) definition of and attitude toward pediatric palliative care; (2) current provision of pediatric palliative care; (3) the support needs of healthcare professionals in the provision of pediatric palliative care; and (4) the role of specialised pediatric palliative care teams. Healthcare professionals expressed openness to pediatric palliative care and reported distinctive needs for support in the care of these patients. The main tasks of specialised pediatric palliative care teams in Switzerland would encompass the coaching of attending teams, coordination of care, symptom control, and direct support of affected families during and beyond the illness of their child. This study indicates the need for specialised pediatric palliative care in Switzerland. Specialized pediatric palliative care teams could have a significant impact on the care of children and families with pediatric palliative care needs. Whether hospices are an option in Switzerland remains unanswered; however, a place to meet other families with similar destinies was emphasized.

Clarke and Quin (2007) elicited the views of the multidisciplinary team regarding their experiences of providing a pediatric palliative care service in Ireland. Fifteen participants, including doctors, nurses, physiotherapists, occupational therapists and social workers participated in focus groups. Three theoretical themes emerged. These included clarity of definition and complexity of engagement, seeking to deliver a palliative care service, and the emotional cost of providing palliative care. Further analysis of these themes points to a work-life experience of skilled and emotional engagement with children, and their parents, in complex processes of caregiving and decision-making. Pediatric palliative care occurs in an environment where parents shoulder a large burden of the care and professionals find themselves working in under resourced-services. The participants in this study provided palliative care for children from all four categories of life-limiting conditions. Although similar experiences of caring were common, there were also some notable differences; for example, the greater level and availability of resources for the provision of palliative care services to children dying of cancer than for children with other life-limiting conditions. Importantly, this mirrors the findings of a previously mentioned study which elicited the views of parents concerning the care provided to children with life-limiting conditions (Price et al., 2012). Furthermore, defining caring work as palliative care appeared to be easier when caring for children with a cancer diagnosis than for children with other life-limiting conditions. In both situations it appeared that the imminence of death and the shortness of the terminal phase of the illness were influential. The FMSF, Paterson's Shifting Perspectives Model of Chronic Illness and Bronfenbrenner's Ecological Theory are pertinent in considering the findings. Using the FMSF and Paterson's Shifting Perspectives Model of Chronic Illness to understand how families manage can provide healthcare professionals with insights on how to work effectively with families. Bronfenbrenner's theory reminds healthcare professionals that sometimes they need to advocate for changes in organisations and systems to best support children with NMLLCs and their families.

McCloskey and Taggart (2010) used semi-structured interviews to explore the occupational stress experienced by eighteen nurses providing CPC in Northern Ireland. In total, four focus groups took place with children's hospice nurses, community children's nurses and children's nurse specialists based in the regional children's hospital. Four core themes emerged: work demands; relationships, maintaining control, and support and roles. Nurses commented on the negative impact that stress was placing on their professional and personal lives. Of particular note were stressors associated with the sub-themes of relationships, emotional demands and ethical conflicts. While the opportunity to develop relationships was seen as satisfying, the absence of such a relationship was perceived as stressful and was often associated with late referral to the service. While many nurses

felt satisfied when they perceived the care delivered was good, they experienced stress when the care they delivered was not to the standard they aspired to. The lack of time to meet a child's wider needs beyond the immediate clinical task was perceived as delivering sub-optimal care. They experienced grief when the child died, mirroring the findings of other research studies (Liben, Papadatou and Wolfe, 2008; McConnell, Scott and Porter, 2016). The study highlighted important issues regarding nurses feeling they had to present in a certain manner when managing grief. Ethical conflicts were a source of stress. Such conflicts were intensified through the nurse's responsibility to advocate for the child through the parents who often did not share the nurse's opinion. On a critical note, the study had a limited response of 24%, which the authors raised as a concern for transferring the findings to the wider area of CPC. The authors defended this by commenting on the robustness of the design and 'fit' with previous studies.

McConnell, Scott and Porter (2016) carried out a mixed-method review to explore the experiences of healthcare professionals who provide end-of-life care to children in order to inform the development of interventions to support them, thereby improving the quality of care for both children and their families. Searches included CINAHL, MEDLINE, Web of Science, EMBASE, PsychINFO and the Cochrane Library. Additional literature was uncovered from searching reference lists of relevant studies, along with contacting experts in the field of paediatric palliative care. The sixteen qualitative, six quantitative and eight mixed methods studies identified included healthcare professionals in a range of settings. The majority of studies were conducted in the United States. Key themes identified rewards and challenges of providing end-of-life care to children, the impact on their personal and professional lives, coping strategies and key approaches to help support staff in their role. Healthcare professionals experienced a number of rewards when providing end-of-life care to children that lead to increased job satisfaction. Challenges included time constraints caused by demanding caseloads and staff shortages; communication difficulties with parents, colleagues and management; and barriers to coordinating care with other agencies. The review described the children's hospice setting as the most optimal work environment when compared to hospital and community settings, referring to the informal and home-like setting, and the priority and time afforded to social, emotional and spiritual aspects of care. A number of studies have focused specifically on the experiences of healthcare professionals caring for children in a children's hospice setting.

### **3.2.1 Caring for children in a children's hospice setting**

Taylor and Aldridge (2017) explored the rewards and challenges of working in a children's hospice in the United Kingdom with an aim to identify staff support and development needs. Thirty-four care team members including medical, nursing and allied health professionals participated in an interview. Seventeen care team members took part in focus groups. Three care team members took part in both the interview and focus group. The participants described the work as emotionally intensive and multi-faceted. 'Getting it right' was identified as a strong motivator and reward, but also a potential stressor. Other factors were identified as both a reward and stressor, including team functioning, the allocation of work, meeting parent expectations, and the environment. Participants identified training needs to help them feel more confident and competent. They expressed concerns about work-related stress mirroring the findings of the previously mentioned studies (McCloskey and Taggart, 2010; McConnell, Scott and Porter, 2016). Informal support from colleagues and group clinical reflection were identified as primary resources. However, opportunities for this were limited. The authors concluded that organisational structures play a key role in ensuring the emotional investment individuals make in delivering quality palliative care is valued, and that appropriate support, development and training for their role in supporting children and their families and their own wellbeing is available.

McConnell and Porter (2017) explored the impact of providing end-of-life care to children on staff within a hospice setting in Ireland. Fifteen care team staff from a children's hospice offering palliative and specialist care to children and young people with life-limiting conditions participated in interviews and a focus group. Despite the sadness of the circumstances that are inevitably part of working life in a children's hospice, staff found many aspects of the work very rewarding. They repeatedly spoke of 'making a difference' as one of the most positive experiences. While staff identified a number of rewarding experiences, it was also clear that providing end-of-life care to children was associated with many challenging experiences. The findings suggest that the hospice setting provides a model of excellence in supporting staff and mitigating challenging aspects of their role, which includes peer/organisational support, and regular ongoing training in key aspects of children's palliative care. Key recommendations for improving their experience included advanced communication training and knowledge sharing with other children's palliative care specialists within the acute setting. The authors identified a number of limitations in relation to the study. The findings consider a single children's hospice, with a small sample size, limiting transferability to other settings. While being open to healthcare assistants, the sample was mostly comprised of nurses, which makes it less representative of all care staff. Nonetheless, many of the findings substantiate

results from research in a range of settings. Studies focusing on the experiences of healthcare professionals caring for children in the community will be discussed next.

### **3.2.2 Caring for children in the community**

A number of studies focused on healthcare professionals' views regarding the delivery of CPC services to children with life-limiting and life-threatening conditions in the community (Pontin and Lewis, 2008; Quinn and Bailey, 2011; Reid, 2013). The participants demonstrated their commitment to consult, coordinate, negotiate, and ultimately deliver the care required by children and families, but against a background of issues relating to the complexity of the role, and the pressure that such work incurs. Pontin and Lewis (2008) explored how community children's nurses deliver services to children with life-limiting, life-threatening and chronic conditions in the United Kingdom. The study generated a number of insights about the sorts of phenomena that contribute to community children's nurses perceptions of workload. The analysis of the interviews identified the mechanisms and strategies they used for managing their work, meeting clients' needs while ensuring that continuity of care and carer was maintained. From their responses to questions, the responsibility relationship and autonomy characteristics of their role were perceived to be a good thing. However, they acknowledged that working in such a way is stressful and provided examples from their everyday working lives. They emphasised the role of support from colleagues as an important way of maintaining and sustaining the responsibility relationships inherent in their work pattern. However the findings may not be generalisable to other contexts. The group of community children's nurses in this project actively manage their caseloads to maintain the continuity of care and carer in a particular model of service delivery.

Reid (2013) undertook a study to elicit the views of children's nurses with regard to the personal, contextual and interprofessional challenges faced when delivering palliative and end-of-life care to children and young people in the community in Scotland. Seven nurses (four children's community nurses and three children's respite nurses) who provide palliative care to one or more child or young person in the home were interviewed. Four themes emerged: service delivery, nurse-family relationships, nurses' grief, funeral rites and bereavement support. One of the main challenges identified by all participants, especially when there was rapid turnover of staff, was initiating and sustaining plans of care across a range of contexts. Maintaining professional boundaries when trying to balance familiarity and emotional involvement with a level of detachment was challenging for nurses. Although participants appeared to be reconciled to child death, all still described strong feelings of fearful anticipation, emotional wrestling and cumulative effects. The funeral appeared to

offer a setting where most nurses could legitimately demonstrate personal grief. Providing post-death bereavement support was considered to be part of their role if requested by the family, although this was not always recognised by managers.

A common thread throughout the literature related to the emotional cost of providing palliative care. Forster and Haiz (2015) explored health professionals' perceptions of bereavement support surrounding the loss of a child in Australia. Semi-structured interviews were conducted with ten health professionals including doctors, nurses and social workers who were directly involved in the care of the dying child and family in seven cases of paediatric death in medical, oncology and paediatric intensive care at a tertiary pediatric hospital. The diagnoses were not revealed. For health professionals, constructions around coping emerged as peer support, personal coping strategies, family support, physical impact of support and spiritual beliefs. The first way that coping was constructed by participants was confiding in peers and seeking peer support. This was the most common coping strategy reported by health professionals. Analysis of the narratives also revealed health professionals' perceptions of their support provision. When they reflected on their ability to provide support to families, they referred to not knowing how parents felt about the support they provided. They also described sometimes muddling through the process due to a lack of educational preparation concerning the best way to provide support. Similar to the findings of the previously mentioned studies the findings indicate that caring and supporting children and families has a pervasive emotional and physical impact on health professionals who need peer support, mentoring and strategies for managing in order to continue to provide this much needed care.

### **3.2.3 Interim comments in relation to the experiences of healthcare professionals caring for children with NMLLCs**

From the literature, it is apparent that the experience of caring for children with NMLLCs is a multi-faceted experience for healthcare professionals. However, many of the studies previously undertaken focused on a single care setting or a particular care environment. Several studies were conducted in a children's hospice setting (Hunter, 2017; McConnell and Porter, 2017; Taylor and Aldridge, 2017). A number of studies were also conducted in community settings (Pontin and Lewis, 2008; Quinn and Bailey, 2011; Reid, 2013). While these studies are informative they provide a distinctive perspective. Additionally, several studies concentrated on the experiences of nurses (McCloskey and Taggart, 2010; McConnell and Porter, 2017; Pontin and Lewis, 2008; Quinn and Bailey, 2011; Reid, 2013). Yet the multidisciplinary nature of CPC has been previously emphasised (Postier, Catrine and Remke, 2018). Children, parents and families often need considerable

investment in time and call for a multidisciplinary action of a highly complex nature. Children are in continuous physical, emotional and cognitive evolution, and this affects every aspect of their care, from the dosage of medication to the choice of methods for communication and support (Benini et al., 2008).

### **3.3 Access**

As previously stated (see Chapter 1) access, in this study, ultimately refers to the ability of families to obtain appropriate and necessary healthcare services for the child and the family unit as a whole. Parents/families of children with life-limiting conditions require early and ongoing support with their child's health and social care, and help to minimise the wider impact on the family (Carter, Edwards and Hunt, 2015; Kirk and Pritchard, 2012; Noyes et al., 2013). Some children and their families need this ongoing support over decades (IHF/LLH, 2013; Liben, Papadatou and Wolfe, 2008; Noyes et al., 2013; Taylor et al., 2010). Many countries, including Ireland have made CPC and support to families a priority (Craft and Killen, 2007; DoHC, 2009; Noyes et al., 2013). The subsequent sections will explore the current literature base surrounding accessing services. A number of studies dealt broadly with the issue of accessing services. These studies will be critiqued first.

#### **3.3.1 Studies dealing broadly with the issue of access to services**

Coad et al. (2015) explored the perceived met and unmet needs of children/young people with life-limiting conditions and their families in the United Kingdom. This study was a pioneering study in the area of CPC commissioned by the UK charity, Together for Short Lives against which numerous other studies are discussed. Fifty-nine adult individuals who were part of fifty-one families participated in semi-structured, in-depth interviews. This included forty-three mothers; eight fathers and the remainder eight were key family members (birth grandparents, foster grandparents and adoptive parents) who play a primary care role for the child or young person. Eighteen children and young people were also interviewed. Thus, this study was both family-centred and child-centred in its approach. In line with the Appreciative Inquiry approach, interviews included participatory arts-based methods to help participants to explore what was good about services (met needs); what could be better about services (unmet needs) and what the ideal for future services would look like. Children's hospice services were perceived as very beneficial to the family members who had used them. Through the opportunity to access children's hospices facilities, parents and carers were given time and space to separate themselves from their normal caring duties. They felt that staff were knowledgeable and compassionate to their specific individualised needs but also provided the specialist skills for children/young people with life-limiting conditions and their

families. This finding was echoed in a number of studies eliciting the views of service users regarding children's hospice services in the United Kingdom (Grinyer, Payne and Barbarachild, 2010; Price, McCloskey and Brazil, 2017). Support within schools for children/young people with life-limiting conditions was very varied but overall was reported to be important in meeting needs. Support for transition between phases of education likewise varied. Services provided through schools (e.g. physiotherapy) which were accessible under one space/area were very useful for families. A single port of call, in many cases, was called for as this was felt that one person could ideally be placed to listen to them and support their needs. A number of families spoke about having a lead discharge nurse in the hospital or hospices and/or a team available that had enabled them to be discharged 'earlier'. Accessibility of support was highlighted as important as was being able to make contact and to receive support out of normal working hours.

Another key finding from the research was parents and carers, especially mothers (who were frequently the main carers), expressed their frustrations about being 'exhausted' especially if 'breaks' in care were not provided (Coad et al., 2015). Numerous studies highlight the impact of providing care to children with life-limiting conditions, including the pervasiveness of fatigue and exhaustion (Collins et al., 2016; Rodriguez and King, 2009; Steele and Davies, 2006). Courtney et al. (2018) describe exhaustion as a pervasive problem for mothers caring for children with life-limiting conditions. In the said study it was attributed to a chronic lack of sleep associated with care requirements. Despite their exhaustion, mothers described the need to remain constantly alert and responsive to their child's health. In the aforementioned study the complex psychological support needs of family members were thought to often be left unmet, because of the focus on treating the affected child/young person (Coad et al., 2015). There were many instances cited when services had not met needs. Somanadhan and Larkin (2016) suggest that there is a consistent shortfall in social, emotional and respite support for families of children with life-limiting conditions. Equipment needs at home such as feeding equipment, disposables such as syringes or dressings, nappies and ventilation equipment were a repeated source of concern and contention for the family carers, especially mothers. The findings from a number of studies have likewise identified major problems in the provision of specialist equipment for families who are caring for children with life-limiting conditions and who are technology dependent at home (Courtney et al., 2018; Weaver et al., 2018).

Participants in the study undertaken by Coad et al. (2015) reported that they had to retell their 'stories' many times to professionals because collaboration within and between service providers was often fragmented. Many of the children and young people frequently experienced an acute

medical crisis and had to be admitted to hospital as an emergency. Here, there appeared to be few systems in place which enabled a child or young person's case history to be retrieved immediately on their admission. There appeared to be a lack of centralised information regarding voluntary/support services. Parents and legal guardians made a heartrending plea for easily accessible information, using information technology systems that were responsive and preferably obtainable from a single port of call. Overall the findings indicated that children and their families felt medical/nursing needs were well met but provision was needed for broader financial, social and emotional support alongside more responsive specialist therapies. Although this study was conducted in one large area of the United Kingdom it has international implications. However, it must be acknowledged that access to palliative care services, in particular hospice services varies within regions (Bergstraesser et al., 2013; Davies, 2005; Grinyer, Payne and Barbarachild, 2010; Price, Jordan and Prior, 2013). This has significant implications such as difficulty in comparing findings. What is new about the study is that the researchers interviewed children, young people and family members about their perceptions of met and unmet needs in terms of palliative care services.

Price et al. (2012) examined the experiences of bereaved parents concerning the care provided to children who died from cancer compared to those who died from a non-malignant condition in Ireland. Twenty-five parents (16 mothers and 9 fathers) talked about the life and death of 16 children. Ten of the children had non-malignant conditions while six had cancer. Although parents' accounts displayed commonalities, key differences were discernible. Typically, parents of children with cancer considered that care at the end-of-life as well-resourced and responsive to their and their child's needs. In contrast, parents of children with non-malignant conditions reported under-resourced and inadequately responsive services. Although both groups of parents called extensively on military metaphors such as 'battle', 'fight' and 'struggle' the focus of their respective energies was different. In the one case the adversary was disease and illness; in the other it was the service providers and service provision. Parents of children with non-malignant life-limiting conditions described expending huge amounts of emotional and physical energy in their 'battle' for adequate service provision, aware of having to struggle with what they saw as two related fronts: bureaucratic ineptitude and inadequate resources. Consequently, frustration and resentment characterised these parents' accounts, particularly in relation to acquiring services to facilitate home care. They highlighted a disorganised and uncompassionate approach on the part of formal service providers, coalescing around the assessment process for eligibility to services. In this context, their experience of children's hospice services, both community and hospice based, was of a 'lifeline'. However,

delayed referral to hospice services featured in the accounts of parents in the non-malignant grouping. Of particular importance for those in the non-malignant group was the availability of respite care, which allowed parents to invest time in family related activity. For parents of children with non-malignant conditions, the infrastructure is less well developed, less easily accessed and less flexible. These inequities are reflected in the ways that parents talk about their experiences of caring, in particular, how parents of children with non-malignant conditions repeatedly talk about having to 'fight' for resources.

Interestingly Dybvik et al. (2011) described how the core category, 'fighting the system,' became the central theme as family members were asked to describe their experiences of giving advanced care to family members dependent on home mechanical ventilation in Norway. A total of fifteen family members were interviewed regarding their experience of giving advanced care to eleven ventilator-dependent individuals (3 children and 8 adults). Based on the extensive experience the family members had, they were mostly focused on describing the continual struggle with the community healthcare services, or 'the system', as they called it. Even though their family member was completely dependent on highly advanced medical procedures and technological equipment, the 'fight against the system' seemed to be the most problematic issue for families. Similarly Somanadhan and Larkin (2016) outlined that families of children with life-limiting conditions experienced difficulties in accessing medical and non-medical services for their children in Ireland. A total of eight parents of children with a range of Mucopolysaccharidosis (MPS) disorders aged from 6 months to 22 years were interviewed. Families described the dynamics of relationships in terms of both family relations, reflecting their immediate family and close friends in a supportive care capacity, and of relations with the healthcare system and services, as a non-relational relationship. They reported that coordination and communication between healthcare services are fragmented. They spoke about spending significant amount of time navigating bureaucracy instead of caring for their sick children. The commonality that exists between these studies and the previously mentioned studies is the requisite for services and support for families of children with NMLLCs. However, families generally reported limitations in the availability and access to care, and wide variation in the accessibility of information and lack of shared knowledge.

In a newly published study Kiernan et al. (2019) explored parents' experiences of services for children with life-limiting neurodevelopmental disabilities in Ireland using mixed methods. The study included a quantitative survey (n = 63) and qualitative interviews (n = 12), with a focus on integrating the findings to identify higher-level insights. Results suggest parents' experiences are

mixed, showing more negative than positive experiences. Access to services was described as problematic, with obstacles, including funding and geographical inequities reflecting the findings of previously mentioned studies (Coad et al., 2015; Price et al., 2012; Somanadhan and Larkin, 2016). Helpful aspects included staff attitudes and relationships and, to a lesser extent, coordinated care. Unhelpful aspects included inconsistencies in the provision of services. The findings suggest some implications for service provision, including the need for a more family-centred approach. However the authors identified a number of limitations in relation to the study. This study was conducted within one service in Ireland. Another limitation is the dominance of mothers' views.

Weaver et al. (2018) explored how the pediatric home nursing shortage translates into a lived experience for families with children with complex medical conditions receiving palliative care in the United States. A total of 38 home health nursing surveys were completed by families receiving pediatric palliative care consultation services at a freestanding children's hospital in the Midwest. The gap in the average number of nursing hours allotted versus received was 40 hours per week per family, primarily during evening hours. Families invested an average of 10 hours per month searching for additional nursing coverage and often resorted to utilizing more than 6 different home nurse coverage personnel per month. Families reported multiple delays to hospital discharges due to inability to find home nursing coverage. Respiratory technology and lack of Medicaid coverage correlated with the gap in home nursing access. Limitations to this study include small sample size and one-site location at a freestanding children's hospital in the Midwest.

### **3.3.2 Access to services at specific points in the care trajectory**

A few studies focused on the issue of access to services for children with life-limiting conditions at specific points in the care trajectory, such as the transition to home or the transition from child to adult services. Many children with complex medical conditions (particularly those children who are technology dependent on tracheostomies or feeding devices) undergo extended hospital stays while awaiting home nursing access for safe discharge from the in-patient setting (Abode et al., 2011; Amin et al., 2015). Brenner et al. (2015) explored parents' perspectives of the transition to home of a child with complex respiratory healthcare needs in Ireland. Fifteen parents participated (12 mothers and 3 fathers), representing the transition to home of fifteen children from a large Children's hospital in the previous 5 years. Although there was no reference to life-limiting it was apparent that the children had very complex medical needs. Additionally, many parents expressed concern regarding the future, suggesting that the future could be very negative to contemplate. Telephone interviews were conducted using open-ended questions. Four key themes which

emerged from the interviews were 'stepping stones: negotiating the move to home', 'fighting and frustration', 'questioning competence' and 'coping into the future'. The theme 'fighting and frustration' in particular resonated with many of the previously mentioned studies (Courtney et al., 2018; Dybwick et al., 2011; Price et al., 2012; Somanadhan and Larkin, 2016). Each of the parents expressed their anger and frustration at the length of time it took to have everything in place so they could take their child home. Many of the parents interviewed raised concern about the quality of care available in the community and in regional hospitals once they had been discharged from the tertiary care centre. Parents spoke at length about their ongoing challenges and their hopes for the future. This included difficulty thinking beyond the present moment, concern for their child coping with their level of ability and the need for a compassionate health service to help parents cope. The use of telephone interviews as opposed to face-to-face interviews could be viewed as a limitation of the study. Telephone interviews are often viewed as a less attractive alternative to face-to-face interviews as they restrict the development of a rapport (Irvine, 2011; Rahman, 2015). The authors of the study however suggest that this was not their experience.

Improvements in care and treatment have led to more young adults with life-limiting conditions living beyond childhood, which means they must make the transition from children's to adult services (Kerr et al., 2017; Moola, 2012). This has proved a challenging process for both young adults and service providers, with complex transition interventions interacting in unpredictable ways with local contexts (Kerr et al., 2017). The timing of transition is considered paramount and must be seen as a continuous process, not a single event (Clarke et al., 2017; Hudsmith and Thorne, 2007). Although transition should be viewed as individual, there should be a plan for the future of the child that begins with the end of paediatric palliative care in mind (HSE, 2018). Doug et al. (2011) undertook a systematic review to evaluate the evidence on the transition process from child to adult services for young people with palliative care needs in the United Kingdom. The bibliographic databases Medline, CINAHL, PsychINFO, British Education Index and the International Bibliography of the Social Sciences from 1995 to 2008 were searched. The authors also searched the reference lists of included studies and sought additional literature through contact with experts in palliative care. Ninety-two papers met the inclusion criteria and were from a variety of countries around the world. The findings demonstrate that there is no evidence of transition services that address 'palliative care' as an overall concept. Transition programs that exist are mainly condition-specific. No long-term outcome data exist to compare the effectiveness/cost effectiveness of the condition-specific or generic transition models. Some consensus exists on what may facilitate or impede

successful transition programs. Transition should be multidisciplinary in nature, individualised in terms of addressing need and navigated with the help of a designated key worker.

### **3.3.3 Access surrounding specific aspects of service provision**

Numerous studies addressed access surrounding specific aspects of service provision such as respite care, hospice or home-based support. Respite is a service that allows carers to take 'a break' and leave their child with a substitute caregiver by formal and informal means (Smith, Graham and Herbert, 2017). This aims to reduce the stress and fatigue that comes with the continuous provision of care (Eaton, 2008; Ling et al., 2015). Respite may also provide an opportunity for the child to increase their own social interactions and vary their activity (Ling et al., 2015). Grinyer, Payne and Barbarachild (2010) elicited the views of service users on their experiences of respite care in a children's hospice in Northern England. Twenty-four service users (11 families) which included: parents, children and young people, siblings, guardians and family carers participated in in-depth interviews. Hence again this study was both family-centred and child-centred in its approach. The findings revealed a lack of choice, practical problems of access and powerless dependency surrounding respite care. There seemed to be little choice about when, how often, and for how long respite care was offered. The duration of the respite care was often very short-sometimes a single night-and the complicated preparations necessary were thought by some to be disproportionate. Parents were willing to endure difficulties and frustrations discussed above because they experienced the respite service as a 'lifeline'. This study does have limitations in that it is based on the evaluation of a single care setting and a relatively small number of participants.

Smith, Graham and Herbert (2017) also elicited the views of service users on their experiences of respite care in Australia. This study was undertaken to clarify the respite needs of parents caring for children with life-limiting conditions by first identifying the respite, both formal and informal, that these families access and then identifying preferences for future respite. Additionally, it was undertaken to understand barriers that families have to accessing the respite they need in caring for their child. Thirty-four families cared for by the Paediatric Palliative Care Service in Queensland were approached to participate in a survey. Twenty surveys were returned. Three of the families reported receiving no respite in the previous 12 months. Families who received respite received a combination of formal respite (a structured care provider) and informal respite (family or friends). Ten of the families reported that they would want the time of respite changed. Barriers to receiving adequate respite included complexity surrounding the care of the child, financial barriers and lack of a respite provider. The authors identified several limitations concerning the study. They suggest

that caution must be exercised when generalising the findings of this study due to the small sample size. Certain groups are under-represented in the sample, such as families with culturally diverse backgrounds and those living in a rural location. For families who are not accessing respite, it is important to clarify whether this is due to their preference or due to a lack of services. The authors suggest that the perceived benefits of respite and whether parents felt they had received a break from care giving in the respite that is provided is also an important area for future research. There is a dearth of evidence on the actual provision of respite care for children with life-limiting conditions. The international literature has established that respite care is insufficient and yet essential in addressing the palliative care needs of children (Eaton, 2008; Grinyer, Payne and Barbarachild, 2010; Ling et al., 2015; Smith, Graham and Herbert, 2017). Parents in the aforementioned studies reported vastly different needs and experiences of respite care (Grinyer, Payne and Barbarachild (2010; Smith, Graham and Herbert, 2017).

Kirk and Pritchard (2012) undertook a study in the United Kingdom to investigate parents' and young people's perceptions of hospice support and identify how support could be improved. A mixed methods design was used. A total of 108 postal questionnaires were returned from parents who had used the services of one hospice in the previous two years and interviews were conducted with 12 parents and 7 young people sampled from the postal questionnaire. Of those who completed the postal questionnaire 37 were bereaved. Families were highly satisfied with the support provided in terms of quality of care; interpersonal qualities of the staff; the individualized, family-focused approach; accessibility of support and involvement in decision-making. This again mirrors the findings of previous studies (Coad et al., 2015; Grinyer, Payne and Barbarachild, 2010; Price et al., 2012; Price, McCloskey and Brazil, 2017). Young people valued the opportunity to meet with other young people and take part in different activities. For parents the provision of a break from caring was the main way in which they were supported, although they felt they wanted more of this form of support. A consistent theme in relation to support for young people and siblings was the need to develop facilities, activities and bereavement support specifically for teenagers/young people. Again, this study does have limitations in that it is centred on a single care setting and a relatively small number of participants.

While parents attach importance to home-based care (Carnevale et al., 2006; Carter and Mandrell, 2013), support services are not always readily available (Heath et al., 2012; Winitzer et al., 2012). Only a minority of families can access regular respite (Grinyer, Payne and Barbarachild, 2010) or comprehensive palliative care (Bradford et al., 2012). Where statutory services are non-existent or

unable to meet demand, gaps occur in support networks (Bradford et al., 2012; Nageswaran et al., 2012). Some gaps are filled by charitable organizations (Carter, Edwards and Hunt, 2015). For example, family support workers provide practical, non-nursing support to the family, such as babysitting, help with shopping and transport to/from hospital. Family support workers are not registered health professionals, but are selected for their skills and experiences relating to children and families. Carter, Edwards and Hunt (2015) explored key aspects of the work of the family support workers in caring and supporting families of children with life-limiting conditions from the families' perspectives in the United Kingdom. This study used a mixed method design. The participants included fifty-five families (12 bereaved) and thirty-nine children aged 2–18 years. Thematic analysis revealed how the family support workers became a presence in families' lives in three main ways: (1) *encompassing and embracing* families through supporting needs and promoting resilience; (2) *befriending and bonding* through developing knowledge, trusting relationships and a sense of closeness; and (3) *accompanying and enduring* by 'being with' families in different settings, situations and crises and by enduring alongside the families. The study demonstrated the fundamental importance of support workers who are able to provide aspects of support that is usually not provided by other services. The authors acknowledged a number of study limitations. They stated that although the number of participants in the study is relatively high when compared to similar studies, a limitation to the study is the low sample size, in relation to the target population and this potentially limits representativeness. The low response rate to the survey reflected a number of challenges with regard to the distribution of questionnaires. Families expressed altruistic reasons for engaging in the study and may also have felt that speaking positively about the service would provide evidence to support service continuation.

The concept of a volunteer service has been advocated for caregivers and siblings of children with life-limiting conditions (Bona, Bates and Wolfe, 2011; Burbeck et al., 2015; Carter and Mandrell, 2013). McGrath (2001) obtained feedback on the need for a volunteer service for families coping with a child with a life-limiting condition in Australia. A short (two page) questionnaire was developed to obtain feedback. Questions were asked about the perceived need for trained volunteers, what activities they could help with, whether the need for them changes over time, obstacles to their participation, and previous experience with volunteers. One-hundred and seven families with a child diagnosed with a life-limiting condition completed the questionnaire. The questionnaire responses indicated that many families have only minimal or no support, and were coping with extraordinary physical, emotional and social demands from the child's condition and treatment. The participants were very positive about the need for a trained volunteer and were

clear about the activities that trained volunteers could provide assistance with. These activities ranged from practical assistance such as babysitting and help with household chores and errands to emotional support. Not all families would be comfortable with a volunteer, and some respondents did outline perceived obstacles to including a volunteer in family activities. For others there were comments about why volunteers would be perceived as unproblematic. Notably, over half of the questionnaires were returned from the Cystic Fibrosis Association. Whilst these children are categorised as life-limiting they have distinct needs and trajectories as compared to other children with life-limiting conditions.

### **3.3.4 Interim comments regarding accessing services**

There is now growing recognition of the needs of infants, children/young people with life-limiting conditions and their families and an increasing understanding of the full breadth and scope of CPC (Derrington, 2015; Hauer and Wolfe, 2014). There are also gaps in the existing literature base. There is limited evidence regarding parents/families views of accessing services when caring for their child. A number of studies focus on specific points in the care trajectory or on specific services such as respite care, hospice or home-based support. Whilst these studies are insightful there is a need to understand parents' views of accessing all aspects of service provision throughout the care trajectory. Indeed, it has been previously highlighted that accessing services can be difficult at critical time points such as the point of diagnosis or recognition of a life-limiting condition, during periods of 'ongoing care' and when the child requires end-of-life care (Noyes et al., 2013; Price et al., 2012; Rallison and Raffin-Bouchal, 2013). Additionally, many of the studies have been conducted outside of Ireland and include a small sample size and one site location. It is still the case that the overall provision of palliative care for children is wholly inadequate in most parts of the world (WHO, 2018). The lack of sustainable funding and problems with coordination of services means that palliative care for children remains patchy and inequitable (Marston and Chambers, 2012). Thus, it is imperative to explore access to services for children with NMLLCs in context.

## **3.4 Decision-making**

Elwyn and Miron-Shatz (2010) describe decision-making as pre-decision deliberation followed by the act of making the determination. Deliberation includes obtaining information and appraising one's own knowledge, imagining alternative outcomes, predicting one's emotional state in the future, and constructing preferences about the decision. Determination is coming to an intention to enact the decision. Decision-making regarding care and interventions for children with NMLLCs is fraught with difficulties but faced regularly by parents and healthcare professionals (Cardona, 2017; Wright et al.,

2009). Decision-making can begin from the point of diagnosis or recognition of a life-limiting condition and can continue through to end-of-life care (Koch and Jones, 2018; Larcher et al., 2015; Toebbe et al., 2013). Parents are frequently asked to make decisions that have significant long-term effects on their child's life, from adding technology (such as a feeding tube or tracheostomy) to considering focusing on supportive care only (Carroll et al., 2012; Klick and Hauer, 2010). Such decisions are complex and influenced by more than simple exchange of information (Cardona 2017; de Vos et al., 2011; Tamburro et al., 2011). Healthcare professionals similarly encounter many challenging and ethical decisions inherent in caring for profoundly medically ill children with life-limiting conditions (Cardona, 2017; de Vos et al., 2011; Lotz et al., 2016). The emotional and physical exhaustion that parents experience as a consequence of their caregiving responsibilities can compound the difficulties associated with decision-making (Nicholson, 2012; Popejoy, 2015). Prognostic uncertainty likewise affects decision-making, making it more difficult for parents and healthcare professionals (Basu and Swil, 2018; Baverstock and Finlay, 2008; Kirschen and Walter, 2015).

The importance of understanding how parents make decisions is necessary to prevent parental regret about decision-making, which can lead to psychological distress, decreased physical health, and decreased quality of life for the parents (Allen, 2014). In addition, understanding how and why parents choose specific treatment and care options is increasingly relevant given the advances in technology and experimental therapies for children with NMLLCs (Amin et al., 2017; Wallis et al., 2011). Shared decision-making is of course advocated in CPC (Santoro and Bennett, 2018). Healthcare professionals have an obligation to broker or participate in the decision-making process (Zimmermann et al., 2016). Accordingly, partnerships between parents and healthcare professionals are critically important in relation to decision-making (Knapp and Contro, 2009). The child's voice is also critical in this partnership (Dunlop, 2008; Nicholson, 2012).

Given the importance of decision-making for children with NMLLCs and their families, surprisingly few studies have addressed this topic and those that do exist have focused primarily on decisions made at end-of-life (Beecham et al., 2017; Geurtzen et al., 2017; Popejoy, 2015; Xafis, Wilkinson and Sullivan, 2015). Yet in several studies, parents of children with NMLLCs identified that decision-making is an integral component of their daily life; requiring a delicate balance alongside managing the day-to-day care of their child and the practicalities of 'normal life' (Erby, Rushton and Geller, 2006). The findings of a study undertaken by Nicholson (2012) revealed that families often encountered errors within the healthcare setting, such as medication errors, surgical complications

or inability of professionals to notice the subtle signs of deterioration in their child's condition. As a result, families made the decision to stay with their child continually during a hospital admission, because of lack of confidence in healthcare professionals. Additionally, healthcare professionals identified that likely deteriorations in the child's condition are often not anticipated or planned for appropriately and children are subsequently given inappropriate treatment, whereas better planning may have resulted in more appropriate outcomes (Mitchell and Dale, 2015).

Koch and Jones (2018) suggest that these parents engage with decision-making that often includes considering mundane, everyday questions not related to end-of-life conversations. This is clearly evident in the aforesaid study (Nicholson, 2012). In a study undertaken in the United States Trowbridge et al. (2017) describe decision-making as a dynamic process for families caring for children with life-limiting conditions. For each family, decision-making was not a single act; each diagnostic test, intervention, or medication was delicately considered based on its potential to help versus potential to harm. Hence it is imperative to understand decision-making over the entire course of the care trajectory, not only at points where parents are choosing between the life and death of their child. However, the findings from these studies provide valuable insight into parental decision-making during times of crisis. The subsequent sections will explore the current literature base surrounding decision-making for families of children with NMLLCs. To commence studies which have investigated factors which assist or impede parental decision-making will be discussed.

### **3.4.1 Factors that affect decision-making**

A number of studies have investigated factors which assist or impede parental decision-making (Atout, Hemingway and Seymour, 2017; Peay et al., 2016; Popejoy et al., 2017). These studies suggest that decision-making and future planning for children with life-limiting conditions is influenced by a number of factors. These include the type of decision, family factors, relational factors and system factors (de Vos et al., 2011; Edwards et al., 2012; Popejoy et al., 2017). Allen (2014) carried out an integrated literature review in the United States to describe possible factors that affect parental decision-making for medically complex children. PubMed, Cumulative Index of Nursing and Allied Health Literature (CINAHL), and PsycINFO were searched using the combined key terms 'parents and decision-making' to obtain English language publications from 2000 to June 2013. The search strategy generated 336 relevant articles based on their titles with 305 articles eliminated after review of abstract. The findings from each of the 31 articles retained were recorded. The critical decisions included continuation or termination of a high-risk pregnancy, initiation of life-sustaining treatments such as resuscitation, complex cardiothoracic surgery, use of

experimental treatments, end-of-life care, and limitation or withdrawal of support. The results of the integrated review suggest that parental decision-making for children with medically complex conditions is impacted by a range of factors, including the type and content of information provided to them as well as the information they sought, the seriousness of the child's illness, whether other treatment options exist, what is best for the child, religiosity and spirituality, parental factors and support. However the majority of studies focused on one homogenous diagnostic group of premature infants and children with complex congenital heart disease. Thus, comparisons across other child illness categories cannot be made.

In any decision-making process, parents/families are invariably influenced by cultural, societal, religious and familial factors unique to the individual (Santoro and Bennett, 2018). Knapp et al. (2014) investigated how decisional conflict varies among racial and ethnic subgroups in parents whose children have a life-threatening illness in the United States. Two hundred and sixty-six surveys were completed by the parents. To measure decisional conflict and its components, the Decisional Conflict Scale was used (O' Connor, 1995). The findings suggest that minority parents report less Effective Decision Making and report less Support in Decision Making compared to white, non-Hispanic parents. Those who identify as Mexican American and Central/South American report having greater Uncertainty in Choosing Options and less Values Clarity as compared to Puerto Rican or Cuban Americans. The findings also suggest that those whose primary language is not English are associated with greater Uncertainty in Choosing Options. Values Clarity was lower for children who were diagnosed with their life-threatening condition at birth as compared to children diagnosed at a later time. This study investigated decision-making in general. The authors did not predicate this study on specific decisions, such as the decision to create an advance directive. They acknowledge that specifying the decision might yield different results both across and within subgroups.

### **3.4.2 Decision-making over the course of the care trajectory**

A small number of studies have focused on particular decisions that families of children with NMLLCs and healthcare professionals encounter in the course of the care trajectory which are not specifically associated with end-of-life care. Ling et al. (2015) conducted a study in Ireland to examine the views and experiences of parents of children with life-limiting conditions on the topic of out-of-home respite care. Utilising multiple, longitudinal, qualitative case study design, the respite needs and experiences of parents caring for a child with a life-limiting condition were explored. Nine families were recruited and followed for two years. A total of 19 in-depth interviews were conducted with

parents. Many interlinking factors influenced their decision regarding the utilisation of out-of-home respite care, including: past experience of in-patient care, and trust and confidence in care providers. The child's age, diagnosis, geographical location and the family's capacity to meet their child's needs were likewise important in influencing their decision. A proposed model for out-of-home respite care has been developed based on the findings. However, this research is limited to the experiences of nine specific families in one country. Parents experiences were explored through case study methodology, and therefore findings cannot be generalised; however cross comparison revealed commonalities within and between cases. The findings from this study mirror the findings from previously mentioned studies which suggest that decisions are often considered in relation to previous decisions, interventions and the child's prognosis rather than occurring as isolated incidents (Mitchell and Dale, 2015; Nicholson, 2012; Rapoport et al., 2013).

Peay et al. (2016) explored clinicians' perspectives and parents' decision-making regarding children's participation in Duchenne muscular dystrophy (DMD) clinical trials in the United States. Eleven clinicians involved in ten trials and fifteen parents involved in six trials were interviewed. Parents described benefit-risk assessments using information from advocacy, peers, professionals, and sponsors. Strong influence was attributed to the progressive nature of DMD. Most expected direct benefit and few considered the possibility of trial failure. Most made decisions to participate before the informed consent process, but none-the-less perceived informed choice with little to lose for potential gain. Clinicians described more influence on parental decisions than attributed by parents. Clinicians felt responsible to facilitate informed consent while maintaining hope. Both clinicians and parents reported criticisms about the informed consent process and regulatory barriers. The primary limitation of the study is that it was retrospective in that parents were asked to think back to their decision-making process. The timing of the deliberation and informed consent varied; for some parents that process occurred relatively close to the date of the interview, while for others it occurred several years in the past. Once a determination to participate is made, it is possible that parents re-frame their perceptions to be consistent with their decision (Jansen, 2014). Additionally, parents interviewed came from a group of early acceptors of clinical trial participation for their children, and therefore their experiences and perceptions may differ from other parents of children with DMD.

The findings do, however mirror the findings of a study employing narrative analysis undertaken in the United Kingdom regarding the reasons parents chose to accept or decline an invitation to enrol their children in clinical research (Fisher, McKeivitt and Boaz, 2011). Sixteen qualitative studies

exploring the experiences of parents living in five countries whose children had a range of health conditions of varying severity were included in the analysis. The health status of the child appeared to influence parents' reasons for participation. Parents whose children had life-threatening conditions often considered they had no choice but to participate and many welcomed the innovation offered through research participation. Such parents also viewed the risks of research less negatively than those whose children were healthy or in the stable stage of a chronic condition.

### **3.4.3 End-of-life decision-making**

In studies where the views of parents have been sought regarding end-of-life decision-making, findings suggest that parents of children with NMLLCs want to be involved in the decision-making process (Beecham et al., 2017; de Vos et al., 2015; Gillam and Sullivan, 2011). However, of particular note is that parents defined involvement in different ways, ranging from having knowledge about the decision to having the decisive say (Gillam and Sullivan, 2011). There are, of course, exceptions to this—rarely, parents prefer to transfer the role of decision-making to physicians (Atout, Hemingway and Seymour, 2017). Nicholson (2012) revealed that involvement in decision-making amongst parents of children with life-limiting conditions related to the presence, or absence, of perceived choice and existed in situations where the child's condition dictated a particular action to be taken, for example an unsafe swallow leading to termination of oral feeding. Even in situations with little perceived choice, the process of agreeing to a treatment was important for parents as it required them to understand the risks and benefits involved and come to their own conclusions that the treatment is necessary. Changes in the child's illness trajectory are viewed as significant by families of children with life-limiting conditions (Wood et al., 2010), and influences the way they view the condition (Bluebond-Langer et al., 2017). It follows that this may factor into the decisions that families make, but this requires investigation (Popejoy et al., 2017). However, parents do not always feel informed or supported in decision-making at end-of-life (Longden, 2012; Meert et al., 2008). Additionally Popejoy (2015) suggests that parents also find it a challenge to verbalise their decisions at the end of their child's life.

Beecham et al. (2017) investigated how parents of children with life-limiting conditions approach and experience advanced care planning in the United Kingdom. Open-ended, semi-structured interviews were conducted with parents of eighteen children; nine children who were currently receiving palliative care services and nine children who had received palliative care and died. Parents reported having discussions and making decisions about the place of care, place of death and the limitation of treatment. Most decisions were made relatively late in the illness and by

parents who wished to keep their options open. Parents reported different levels of involvement in a range of decisions; many wished to be involved in decision-making but did not always feel able to do so. This study revealed that parents' views may change over time, and it is important to allow them to keep their options open. Additionally parents' approach to decision-making varies by the type of decision required. The primary strength of this study, which was also acknowledged by the authors, was the inclusion of parents of children with a range of life-limiting conditions, both deceased and alive, and not restricting the study to decisions about predefined options. The sample was limited to the families of eighteen children, and in most cases, only the child's mother participated in the interview.

Parents repeatedly suggest that honest and timely discussions with physicians are critically important with regard to decision-making for their children who have life-limiting conditions (Longden, 2012; Meert et al., 2008). Xafis, Wilkinson and Sullivan (2015) propose that the timing of introduction of discussions may impact on perceived choice in end-of-life decision-making. The extent to which families were given sufficient time to consider the decision regarding technological support and whether the decision could be predicted in advance appeared to be important in a previously mentioned study (Nicholson, 2012). And yet preference for the timing of the introduction of discussions appears to be individual for each family (Dellon et al., 2010; Edwards et al., 2012; Jackson, Cheater and Reid, 2008). Physicians recommend that discussions should occur early around the time of diagnosis or during a period of stability (Basu and Swil, 2018; Donnelly et al., 2005). However, in reality discussions with parents/families regularly occurred late in the care trajectory after multiple, acute, severe deteriorations (Basu and Swil, 2018; Beecham et al., 2017; Edwards et al., 2012).

Xafis, Wilkinson and Sullivan (2015) carried out a meta-analysis of predominantly empirical research pertaining to features which assist or impede parental end-of-life decisions to provide insight and guidance regarding the development of written resources for parents in Australia. Studies relating to critically ill children and new born infants published between 1990 and 2012 were included. The bibliographic database Scopus was used. The initial search produced 487 publications. Additional papers were identified by hand searching the personal libraries of the authors, and reference lists of relevant articles. Following review of the title and abstract 87 papers were retrieved and read in full text. After the papers were scanned for relevance, the publications were further reduced to 58 for inclusion in the meta-analysis. The most prominently cited needs in the literature related to numerous aspects of information provision; the quantity, quality, delivery, and timing of information

and its provision impacted not only on parents' ability to make end-of-life decisions but also on their emotional wellbeing. The meta-analysis supports the value of written materials, as these provide guidance for both parents and healthcare professionals in pertinent content areas. In response to the complexity associated with decision-making for children with life-limiting conditions a number of decision aids have been developed such as a handbook and web-based resource (Caring Decisions) particularly for parents facing end-of-life decisions for their child. Several studies ascertained parents and healthcare professionals' views on these decision aids (Delany et al., 2017; Xafis et al., 2015). The majority of parents and healthcare professionals viewed them as beneficial. All of the studies were conducted in Australia.

A number of studies have focused on end-of-life decision-making in the pediatric intensive care unit (PICU) (Dryden-Palmer et al., 2018; Launes et al., 2011; Sayeed et al., 2012). Michelson et al. (2013) suggest that parents of children facing the possibility of death in the PICU encounter challenging medical decisions such as pursuing high-risk therapies associated with uncertain outcomes; withdrawing or limiting life sustaining treatments; and organ donation. Parents also face non-medical choices such as deciding: whether to remain with their child during procedures; what to tell their child/sibling(s); and whether asking difficult questions may alienate clinicians. Michelson et al. (2009) undertook a study in the United States to explore factors described by parents in the PICU as important/influential if they were to consider withdrawing life-sustaining therapies. One-to-one semi-structured interviews were conducted with seventy parents whose child was admitted to the PICU for more than 24 hours to up to 1 week. Parents described not having any other treatment options as important/influential if they were to consider withdrawing life-sustaining therapies. The main study limitation stems from asking parents hypothetical questions. Parents' reactions when confronting imminent end-of-life decisions could differ. Considering that more than ninety percent of children admitted to the PICU have survivable conditions, most parents interviewed did not face end-of-life decisions for their children at the time of interview. Thus, these findings cannot be generalised to parents/families of children with NMLLCs. Additionally, many of the studies investigating decision-making in the PICU explored the views of healthcare professionals only (Dryden-Palmer et al., 2018; Lee and Dupree, 2008; Michelson et al., 2013; Mitchell and Dale, 2015).

#### **3.4.4 Shared decision-making in end-of-life decisions**

Caeymaex et al. (2013) suggest that shared decision-making in end-of-life decisions may decrease parental grief in parents. In addition, parents who believe that they have been involved and even that they have taken final responsibility for a decision that has resulted in the death of their child are

mostly not negatively affected by this. Feelings of guilt or regret are no more frequent for them than for parents who have not been decision-makers (Gillam and Sullivan, 2011). However, only one study used a valid and reliable instrument to measure decisional conflict and regret with regard to end-of-life decision-making amongst parents of children with life-limiting conditions (Geurtzen et al., 2017). In the said study Geurtzen et al. (2017) evaluated decisional conflict and regret among parents in the Netherlands regarding the decision on initiating comfort or active care in extreme prematurity. An online survey was used. The survey contained a Decisional Conflict and Decision Regret Scale and was administered nationwide. Sixty-one surveys were returned. The findings revealed little decisional conflict and no decision regret among parents regarding decision-making at twenty-four weeks gestation. On a critical note, the study had a limited response of 27%, which the authors raised as a concern for transferring the findings to the wider area of CPC. In addition, whilst the infants in this study are categorised as life-limiting they have distinct palliative care needs and trajectories. Consequently, these findings cannot be generalised to the entire population of children with life-limiting or life-threatening conditions.

Significantly Rapoport et al. (2013) explored the experiences of bereaved parents when a decision had been made to forgo artificial nutrition and hydration during end-of-life care for their child in the United States. This was a qualitative study using in-depth interviews with eleven parents whose children died after a decision to forgo artificial nutrition and hydration. All parents were satisfied with their decision to forgo artificial nutrition and hydration and believed that their child's death was generally peaceful and comfortable. The child's perceived poor quality of life was central to the decision to forgo artificial nutrition and hydration, with feeding intolerance often contributing to this perception. Despite overall satisfaction, all parents had doubts and questions about the decision and benefited from ongoing assurances from the clinical team. This study has several potential limitations. The findings are limited to these eleven parents and represent retrospective perceptions of their decision to forgo artificial nutrition and hydration. Thus, they may not be generalisable. This study only examined the perceptions of parents who opted to forgo artificial nutrition and hydration. The authors commented that future studies are needed to investigate the experience of parents who chose not to forgo artificial nutrition and hydration.

de Vos et al. (2011) undertook a national cross-sectional study in the Netherlands with 136 paediatric specialists (including oncologists, neurologists, neurosurgeons and metabolic paediatricians) to investigate how they reach end-of-life decisions, how they involve parents, and how they address conflicts. The respondents differed considerably in how they involved parents in

end-of-life decision-making. Half of the respondents choose to inform parents and subsequently, ask for their permission to discontinue treatment, an approach that can be best characterised as shared decision-making. A quarter of the respondents choose to merely inform parents and thus protect them from feeling co-responsible and the remaining quarter of respondents choose to inform parents and allowed them to have the decisive voice. Conflicts within medical teams arose as a result of uncertainties about prognosis and treatment options. Most conflicts with parents arose because parents had a more positive view of the prognosis or had religious objections to treatment discontinuation. All conflicts were eventually resolved through a combination of strategies.

Likewise Lotz et al. (2015) undertook a descriptive qualitative study in Germany to investigate the attitudes and needs of healthcare professionals regarding advanced care planning for children. Seventeen semi-structured interviews were conducted with healthcare professionals caring for severely ill children/adolescents, from different professions, care settings, and institutions. All professional stakeholders perceived advanced care planning as beneficial. The findings also reveal that perceived problems with advance care planning relate to professionals' discomfort and uncertainty regarding end-of-life decisions and advance directives. Conflicts may arise between physicians and non-medical care providers because both avoid taking responsibility for treatment limitations according to a minor's advance directive. Nevertheless, advance care planning is perceived as helpful by providing an action plan for everyone and ensuring that patient/parent wishes are respected. Important requirements for advance care planning were identified as follows: repeated discussions and shared decision-making with the family, a qualified facilitator who ensures continuity throughout the whole process, multi-professional conferences, as well as professional education on advance care planning.

### **3.4.5 Interim comments regarding decision-making**

There is a significant literature base regarding the decision-making experiences of parents/families and healthcare professionals caring for children with NMLLCs. However, the predominant focus has been on specific care decisions such as decisions surrounding experimental treatments (Allen, 2014; Peay et al., 2016) or decisions surrounding advanced care planning (Beecham et al., 2017; Heckford and Beringer, 2014; Popejoy, 2015). Clearly the main focus has been on decisions made at end-of-life (Allen, 2014; Beecham et al., 2017; de Vos et al., 2015; Dunlop, 2008; Geurtzen et al., 2017; Gillam and Sullivan, 2011; Popejoy, 2015; Xafis, Wilkinson and Sullivan, 2011). Although these studies are informative, additional information is required to provide an insight into decision-making regarding the care and management of children with NMLLCs throughout the care trajectory. The

majority of the studies undertaken used a qualitative methodology (Atout, Hemingway and Seymour, 2017; Beecham et al., 2017; Nicholson, 2012; Peay et al., 2016; Popejoy, 2015). Very few studies adopted a quantitative approach (Geurtzen et al., 2017; Knapp et al., 2014; Tamburro et al., 2011), and none of the studies adopted a mixed methods design. Mixed methods design, in which qualitative and quantitative methods are combined can be valuable, where the complementary strengths of each approach can yield greater insight than either approach alone (McKim, 2017). The evidence from the review supports the need to measure decisional conflict for this cohort of parents and families. Parents/families encounter many decisions related to treatment and care options in which decisional conflict may arise (Geurtzen et al., 2017; de Vos et al., 2011; Lotz et al., 2016; Unguru, 2012; Wright et al., 2009). Decision-making for children with NMLLCs is often context dependent (Atout, Hemingway and Seymour, 2017). Several studies were conducted in the United States (Allen, 2014; Cardona, 2017; Donnelly et al., 2005; Edwards et al., 2012; Knapp et al., 2009; Knapp et al., 2014; Peay et al., 2016; Tamburro et al., 2011), and the United Kingdom (Dunlop, 2008; Nicholson, 2012; Popejoy, 2015; Popejoy, 2017). Hence the findings may not be transferable to the Irish context. Yet policy documents in both the Republic of Ireland and Northern Ireland allude to the importance of choice (DoHC, 2009; DoH, 2016). Although the child's voice has been rarely represented in the literature (Nicholson, 2012; Tamburro et al., 2011), the child's best interests were at the centre of all care decisions concerning parents (Beecham et al., 2017; Ling et al., 2015). Additionally the child and family's best interests were at the centre of all decisions concerning healthcare professionals (Cardona, 2017; Donnelly et al., 2005; Edwards et al., 2012; Lotz et al., 2016). This reflected a literature base that was both child and family-centred.

### **3.5 Rationale for the present study**

The integrative literature review has illuminated the challenging and multidimensional nature of caring for children with NMLLCs, both from the perspective of parents and healthcare professionals. While many NMLLCs are extremely rare, their impact is disproportionately great, not only for the children themselves, but on their families and healthcare professionals supporting them (Malcolm et al., 2012; Spathis et al., 2012). Their care is provided in most cases at home by the family over long periods, sometimes over many years (IHF/LLH, 2013; Liben, Papadatou and Wolfe, 2008; Taylor et al., 2010). The prolonged nature of these conditions results in a complex pattern of crises followed by periods of relative stability, necessitating differing levels of services at different stages (Price et al., 2011; Price, McCloskey and Brazil, 2017; Wood et al., 2010). Thus, parenting a child with a NMLLC can lead to emotional and practical chaos for the family, both in the short and longer term.

This means that healthcare professionals and service providers need to be able to recognise changing levels of need and respond accordingly with services that are appropriate.

It is widely accepted by healthcare professionals working with children that palliative care falls into three stages (Together for Short Lives, 2018). These include the point of diagnosis or recognition of a life-limiting condition, during periods of 'ongoing care' and when the child requires end-of-life care. However, despite development of services across countries, accessing services by families, who are central to the ethos of care, can be difficult, particularly during critical time points (Noyes et al., 2013; Price et al., 2012; Rallison and Raffin-Bouchal, 2013). As CPC services develop, it is important to ascertain if the services are meeting the needs of children and parents. Therefore, it is essential to engage with families who are currently using CPC services and ascertain their views and experiences of service provision. Additionally, representing the views of service users is valuable to provide an insight into the reality of their lives. Furthermore, the views and experiences of parents are pertinent in light of the common goal of palliative care to improve quality of life for children and families. It is similarly essential to explore the experiences of healthcare professionals who are caring for children with NMLLCs in order to provide an in-depth understanding of their experiences including their experience of service provision. This will allow for a more comprehensive picture of current palliative care practice.

Decision-making about interventions for children with NMLLCs can begin from the point of diagnosis or recognition of a life-limiting condition and can continue through to end-of-life care. There is overwhelming agreement in the literature that both families and healthcare professionals consider that decision-making is a crucial element in terms of care for children with life-limiting illnesses and conditions (Beecham et al., 2017; Eden and Callister, 2010; Gilmer et al., 2013). In addition, shared decision-making among families and healthcare professionals is advocated in CPC (Caeymaex et al., 2013; Knapp et al., 2010). Given the importance of decision-making for children with NMLLCs, and the emphasis on shared decision-making, it is also critical to explore decision-making regarding the care and management of children with NMLLCs from the perspective of parents and healthcare professionals. The integrative literature review also highlights gaps in the existing literature base. The present study will thus endeavour to address the gaps.

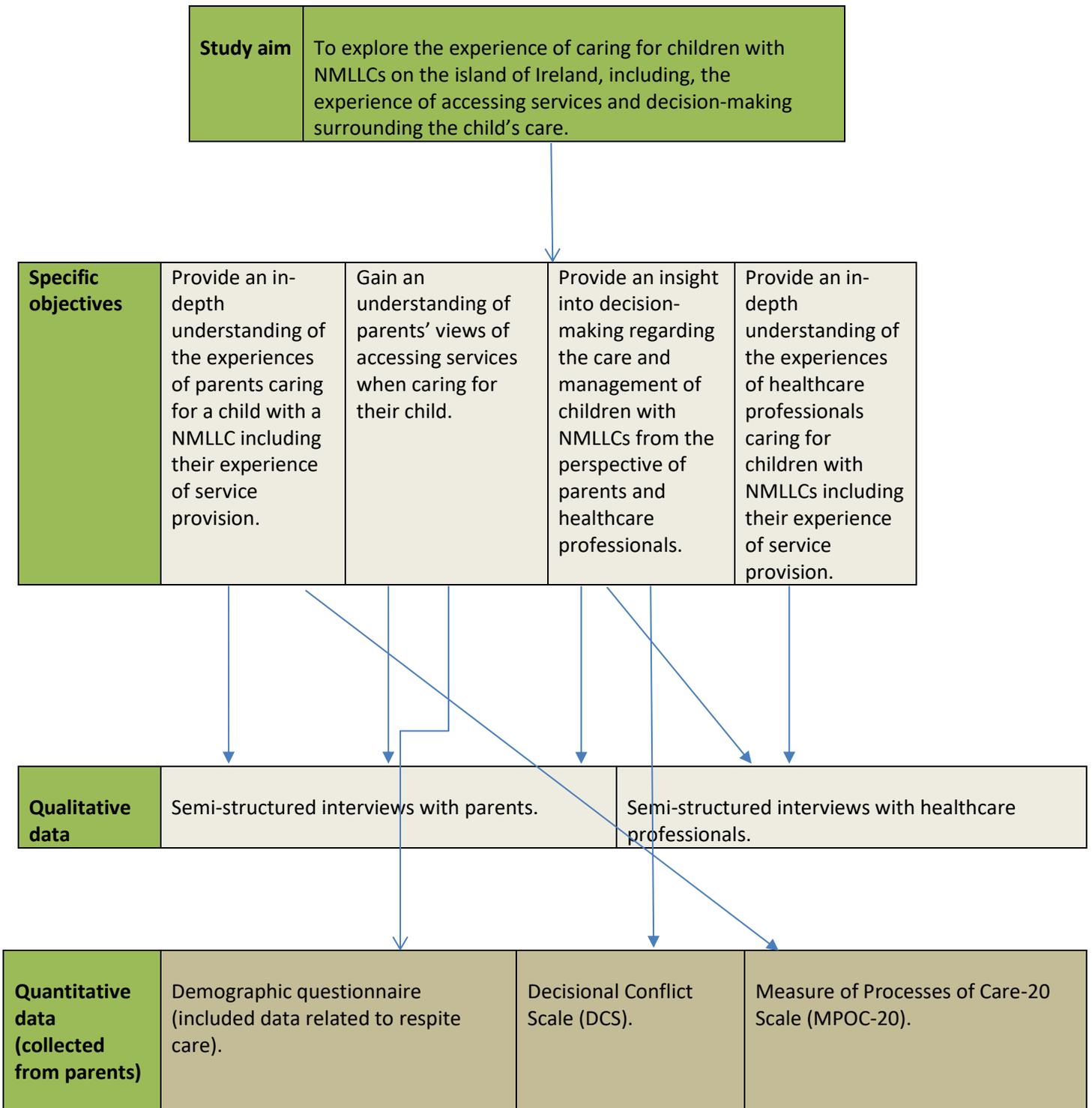
### **3.6 Study aims and objectives**

The aim of this study is to explore the experience of caring for children with NMLLCs on the island of Ireland including, the experience of accessing services and decision-making surrounding the child's care. Specific objectives include to:

- Provide an in-depth understanding of the experiences of parents caring for a child with a NMLLC including their experience of service provision.
- Gain an understanding of parents' views of accessing services when caring for their child.
- Provide an insight into decision-making regarding the care and management of children with NMLLCs from the perspective of parents and healthcare professionals.
- Provide an in-depth understanding of the experiences of healthcare professionals caring for children with NMLLCs including their experience of service provision.

The aims and objectives are diagrammatically represented in Figure 4 and reference is made as to how the mixed methods design is adopted to fulfil this. Greater clarity with regard to this will be provided in Chapter 4.

**Figure 4: Study aims and objectives**



### **3.7 Concluding comments**

This chapter has presented a critical review of the literature related to, but not specifically confined to, the population of this study. This literature review has made clear the challenging and multidimensional nature of caring for children with NMLLCs, both from the perspective of parents and healthcare professionals. The experience of caring for children with NMLLCs has a significant impact on parents and healthcare professionals alike. 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. The majority of the studies were conducted outside of Ireland, where services and care provision differ for these children (Collins et al., 2016; Lazzarin et al., 2018). Additional information is required to provide an insight into decision-making regarding the care and management of children with NMLLCs throughout the care trajectory. There were methodological weaknesses found in the majority of the studies ranging from a single care setting, small sample sizes and low response rates (Dybwik et al., 2011; Hunter, 2017; Smith, Graham and Herbert, 2017). As previously indicated this study will endeavour to address the gaps in the evidence.

## Chapter 4: Methodology

### 4.0 Introduction

This chapter presents an overview of the methodological approach adopted, namely a mixed methods design. The philosophical paradigm of pragmatism that underpins mixed methods research is considered. The rationale for choosing a mixed methods approach and the justification for adopting the concurrent embedded mixed methods design in this study is explained. The advantages and disadvantages associated with employing a mixed methods research design are considered. The method of sampling is identified and the inclusion criteria are outlined. Details regarding the process of recruitment are described in addition to the sample size and participant characteristics. The methods of data-collection are presented and the ethical issues related to the study are addressed. Finally the approach to data analysis and integration is discussed.

### 4.1 Research design

A mixed methods research design was used in this study. In recent years, mixed methods research has become increasingly popular in health services research, allowing a broader deeper understanding of complex human phenomena (Doyle, Brady and Byrne, 2016; Halcomb and Hickman, 2015; O' Cathain, Murphy and Nicholl, 2007). A mixed methods research design is one in which the researcher collects, analyses and integrates both quantitative and qualitative data in a single study (O' Cathain, Murphy and Nicholl, 2010; Onwuegbuzie and Leech, 2006). Its central premise is that the use of quantitative and qualitative approaches, in combination, provides a better understanding of research problems than either approach alone (Creswell and Clark, 2017). Mixed methods research combines quantitative and qualitative research methods which are based on two different paradigms. Quantitative research is based on the positivistic paradigm, that ontologically speaking believes that there is a 'real' reality that can be apprehended, and epistemologically believes that knowledge is objective and that there is a universal truth to findings in empirical studies (Lincoln and Guba, 1985). Qualitative research is based on a new paradigm of science, sometimes referred to as the 'metaphysical paradigm' as it combines a number of different ontologies and epistemologies (Morgan, 2007), which tends to be constructionist in nature, believing reality is relativistic that is, individuals create reality from different ideological, social and personal positions (Cornish and Gillespie, 2009). Both of these viewpoints have been criticised. Positivism has been criticised for prioritising a single form of knowledge as 'true' and therefore ignoring other

alternative forms of knowledge (Cornish and Gillespie, 2009). Constructionism's critics argue that knowledge that is constructed from ideological, social and personal position is simply wrong (Prior, 2003) and as such is not morally defensible when considering the responsibilities of science and research. Recently, however, the concept of pragmatism has been proposed to bridge the gap between the two methods and their paradigms (Bryman, 2016).

#### **4.1.1 Pragmatism**

Mixed methods research advocates a pragmatic approach, which values both subjective and objective knowledge and supports the use of diverse approaches to research on the basis of what works (Creswell and Clark, 2017). The focus is on the consequences of research, on the primary importance of the question asked rather than the methods, and on the use of multiple methods of data collection to inform the problems under study (Creswell and Clark, 2017; Fetters, Curry and Creswell, 2013). Pragmatism is, as the name suggests, practical: whatever works to arrive at good evidence is appropriate (Polit and Beck, 2018). Pragmatist researchers reject a forced choice between the traditional postpositivists' and constructivists' modes of inquiry (Polit and Beck, 2018). The collection of quantitative and qualitative data is seen as compatible and is thought to provide a more complete picture as compared to one data collection strategy alone (Creswell and Clark, 2017). However, the different epistemological and ontological assumptions and paradigms associated with quantitative and qualitative research have had a major influence on discussions on whether the integration of the two is feasible let alone desirable (Morgan, 2007; Wiggins, 2011; Yilmaz, 2013). Proponents of mixed methods research suggest that the purist view, that quantitative and qualitative approaches cannot be merged, poses a threat to the advancement of science (Onwuegbuzie and Leech, 2005) and that while epistemological and ontological commitments maybe associated with certain research methods, the connections are not necessary deterministic (Bryman, 2016). In keeping with the pragmatic theoretical stance, a practical perspective was embraced. The rationale for choosing it for the present study is explained in the next section.

#### **4.1.2 Rationale for using a mixed methods design**

Before making a decision to adopt a mixed methods approach a number of approaches were considered and subsequently rejected. Consideration was given as to whether a quantitative approach or a qualitative approach alone would be most suitable. A vital component of this research was to explore the experiences of parents and healthcare professionals caring for children with NMLLCs and represent their views regarding palliative care service provision. It was clear that these issues could not be adequately addressed using quantitative measures alone. The subjective

voices of participants are not directly heard in quantitative research (Creswell and Clark, 2017). A qualitative approach was considered more appropriate as qualitative designs address the human experience by exploring attitudes, reactions and perceptions (Houser, 2017). However, quantitative data could be used to provide an enhanced understanding of specific concepts, such as decisional conflict and parents' perceptions regarding the extent to which the services they and their child receive are family-centred. Thus, both qualitative and quantitative methods had important roles to play in the achievement of the goals of the study. Therefore a mixed methods design was chosen. Adopting a mixed methods approach allows the researcher to capitalise on the strengths and offset the weaknesses of both quantitative and qualitative research (Bryman, 2016; Fetters, Curry and Creswell, 2013). For instance, quantitative research is typically weak in understanding the context or setting in which people behave, something that qualitative research makes up for (Doyle, Brady and Byrne, 2016; Johnson and Onwuegbuzie, 2004; Yilmaz, 2013). On the other hand, qualitative research is seen as deficient because of the potential for biased interpretations made by the researcher, whereas, quantitative research does not have this weakness (Creswell and Clark, 2017; Doyle, Brady and Byrne, 2016; Yilmaz, 2013). The purpose of using this mixed methods approach was complementarity, that is, that each component addressed a different aspect of the question (Creswell and Clark, 2017).

The qualitative component was used to provide a descriptive rather than statistical account of the experiences of parents and healthcare professionals caring for children with NMLLCs and CPC service provision. The quantitative component was used to provide an enhanced understanding of decision-making and family-centred care. The addition of a quantitative component added a more detailed understanding than could be gleaned from a single perspective (Bryman, 2016; Halcomb and Andrew, 2015; Halcomb and Hickman, 2015). Moreover the quantitative component added insights that may have been missed if a qualitative approach alone was used (Johnson and Onwuegbuzie, 2004). This added to the completeness of the overall study.

#### **4.1.3 Concurrent embedded design**

There are several different typologies of mixed methods designs, including concurrent convergent, sequential explanatory, sequential exploratory, embedded and a multiphase design (Bryman, 2016). A concurrent embedded research design was used in this study. This is a type of mixed methods design whereby the quantitative and qualitative data are collected simultaneously with one dataset labelled primary and the other secondary (Hesse-Biber, 2010). The premises of this design are that a single data set is not sufficient, that each type of question requires different types of data (Creswell

and Clark, 2017). The supplemental data are collected to enhance the overall study as suggested by Creswell and Clark (2017). Semi-structured interviews were used to collect qualitative data from parents and healthcare professionals. The interviews with parents were used to explore their experiences of caring for their child together with their experiences of CPC service provision, including the particular issue of access to services and decision-making surrounding their child's care. The interviews with healthcare professionals were used to explore their experiences of caring for children with NMLLCs including their experience of service provision and decision-making. The use of semi-structured interviews enabled these issues to be explored in detail. As this was an exploratory study the qualitative data were given priority, thus the data collected from the semi-structured interviews were the primary source of data collected.

The quantitative data were the secondary source of data collected. Two standardised measures and a demographic questionnaire were used to collect quantitative data from the parent participants. The standardised measures assessed decisional conflict in parents and also measured parents' perceptions of the extent to which the healthcare services they and their child received were family-centred. The demographic questionnaire was used to gather contextual information regarding the child and family. The quantitative data were collected as there was a compelling argument for some quantitative evidence in order to demonstrate a measurable quantitative aspect to this topic also. For example, the quantitative data could be used to provide additional valuable information about the relative strengths and weaknesses of current CPC service provision. The embedded design was therefore appropriate as the researcher had different questions that required different types of data to address the primary purpose of the study (Creswell and Clark, 2017). Quantitative data were collected from parents only. The views of healthcare professionals were adequately addressed using semi-structured interviews alone. For example, decision-making in the context of CPC service provision was fully explored in the interviews with healthcare professionals whereas an enhanced understanding of the immensely challenging personal decisions that parents encounter was required. This was viewed as important as parents of children with NMLLCs have an imperative role to play in decision-making and regularly face decision-making dilemmas. The quantitative component could be used to measure important aspects of parental decision-making, such as the attitudes and practices of individual healthcare professionals.

#### **4.1.4 Advantages and disadvantages of using a concurrent embedded design**

Despite its considerable advantages as an approach, there are a number of disadvantages associated with using a concurrent embedded design. Compared to quantitative and qualitative research

approaches, mixed methods research is still relatively new (Doyle, Brady and Byrne, 2016; Tariq and Woodman, 2013). Furthermore, mixed methods research is time-consuming and resource intensive (Johnson and Onwuegbuzie, 2004; Tariq and Woodman, 2013). However, this mixed methods design enabled the researcher to collect the data during one data-collection phase. Additionally, within a mixed methods study, there is a requirement to be familiar with both qualitative and quantitative methods independently and how to integrate these methods appropriately to achieve good study outcomes (Doyle, Brady and Byrne, 2016). In general, most researchers are only trained in one form of research inquiry (Doyle, Brady and Byrne, 2016). One of the key challenges within mixed methods research is the successful integration of qualitative and quantitative data during analysis and interpretation (Tariq and Woodman, 2013). Johnson, Onwuegbuzie and Turner (2007) suggest that integration is critical in the context of mixed methods research. However, it is often overlooked in research studies (Creswell and Clark, 2017). Furthermore, it can be unclear how to resolve discrepancies that arise in the interpretation of the findings (Creswell and Clark, 2017). Despite the fact that there is the challenge of uncovering conflicting findings, consistency can be restored through data integration and with the acknowledgement of the complexity of the phenomenon under investigation (Fetters, Curry and Creswell, 2013; Wagner et al., 2012).

## **4.2 Sampling**

Purposive sampling was used for the recruitment of participants. Purposive sampling involves explicitly selecting participants who are likely to generate appropriate and useful data (Green and Thorogood, 2018). It is based on the belief that researchers' knowledge about the population can be used to hand-pick sample members (Polit and Beck, 2018). This method of sampling was chosen for a number of reasons. Firstly, the identification of potential participants for the study was extremely complex. Parents who have a child with a NMLLC who access palliative care services are potentially a small population. In addition, there is no national database to aid identification and selection of possible participants. Furthermore, the identification of parents is further complicated by the fact that children are cared for in a variety of settings. Given these limitations, and the qualitative emphasis within the mixed methods approach of this research, purposive sampling was used to identify parents most capable of providing data on their experiences. Additionally, parents were asked to reflect on the healthcare professionals that they regularly engaged with in the course of care and to identify the professionals that they felt represented their 'key contact'. The parents were therefore placed at the centre of the entire process. It was not the named individual that was important but rather their professional role in CPC service provision. For instance, almost all of the parents identified a paediatrician as a healthcare professional that they regularly engaged with in

the course of care. It was therefore pertinent to include a paediatrician in the overall sample. The inclusion criteria for parents and healthcare professionals are outlined below.

#### **4.2.1 Inclusion criteria**

**Group A:** Parents of children with NMLLCs on the island of Ireland. The parents included mothers and fathers over eighteen years from a wide socio-demographic spread. Fluency in the English language was required.

**Group B:** Healthcare professionals experienced in the care of children with NMLLCs. In order to gain quality data, it was considered necessary that the healthcare professionals would have a minimum of one-year experience in caring for children with NMLLCs on the island of Ireland.

#### **4.2.2 Recruitment of parent participants**

An Advisory Group was established to oversee the implementation of the research. The Advisory Group comprised of healthcare professionals involved in the assessment and provision of CPC and academics involved in CPC research on the island of Ireland. The Advisory Group played a key role in advising re recruitment of parents. Following consultation it was recommended that recruitment should encompass parents from a wide geographical remit on the island of Ireland to include urban and rural locations. It was also suggested that recruitment should encompass a Children's hospital, a paediatric unit within a regional hospital, a respite facility and a children's hospice. Following discussion with members of the Advisory Group and the supervisory team a decision was made to include five organisations in the study. These included a Children's hospital, two paediatric units within regional hospitals, a respite facility and a children's hospice. The organisations were situated in Ireland (two urban and two rural locations) and one situated in the north which covered a wide geographical remit. Recruitment of parents took place through gatekeepers at each organisation. The researcher liaised with the gatekeepers identified from each organisation to recruit parents who met the inclusion criteria of the study. In all organisations the gatekeepers were experienced Children's nurses who were involved in the delivery of care to children with NMLLCs. It was pertinent that the gatekeepers knew the parents as they could seek representation from a wide socio-demographic spread. This was also important as the gatekeepers had an existing relationship with the parents and could therefore introduce the study in a sensitive and timely manner.

The gatekeepers were asked to distribute an Information pack to parents which contained a Letter of invitation (Appendix A) and Information regarding the study (Appendix B). The parents were invited to return an Expression of Interest slip (Appendix C) to the researcher in a stamped

addressed envelope provided, if they were interested in participating in the study. By returning the Expression of Interest slip directly to the researcher it ensured that participation was entirely confidential. The researcher contacted the parents who indicated that they were interested in partaking by telephone. The researcher provided additional information and answered any questions that arose. The interview date and time was then scheduled. Parents were advised that they could phone or text 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. Parents were contacted by text the day before an interview to remind them of the interview.

### **4.2.3 Recruitment of healthcare professionals**

The Advisory Group likewise played a key role in advising re recruitment of healthcare professionals. They suggested that recruitment of healthcare professionals should be guided by parents. As previously mentioned, parents were asked to reflect on the healthcare professionals that they regularly engaged with in the course of care and to identify the professionals that they felt represented their 'key contact'. Four main groups were identified including nurses, social workers, doctors and psychologists. Parents repeatedly identified nurses as key. Two methods of recruitment were used. The researcher, organised information sessions in the five aforementioned organisations for healthcare professionals who were involved in the assessment and provision of care to children with NMLLCs to inform them about the study. These information sessions were targeted for particular groups of healthcare professionals who were specifically identified by parents as representing their 'key contact'. At these information sessions Information packs were distributed which contained a Letter of invitation (Appendix E) and information regarding the study (Appendix F). The healthcare professionals were invited to return an Expression of Interest slip (Appendix G) to the researcher in a stamped addressed envelope provided, if they were interested in participating in the study. The researcher contacted the healthcare professionals who indicated that they were interested in partaking by telephone. The researcher provided additional information and answered any questions that arose. The interview date and time was then scheduled. The second method of recruitment involved contacting healthcare professionals by posting the aforementioned Letter of Invitation and information regarding the study. The healthcare professionals were asked to return an Expression of Interest slip to the researcher. Similarly the researcher contacted the healthcare professionals who indicated that they were interested in partaking by telephone and the interview date and time was scheduled. Additionally, healthcare professionals were advised that they could phone or text 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. Healthcare professionals were contacted by email the day before an interview to remind them of the interview.

#### **4.2.4 Sample size and participant characteristics**

An important stage in planning a study is determining how large a sample size may be required, however, current guidelines for thematic analysis are varied (Fugard and Potts, 2015). Hagaman and Wutich (2016) outline that sixteen or fewer interviews were sufficient to identify common themes. Braun and Clarke (2013) categorise suggestions by the type of data collection and the size of the project. Another approach is to investigate until data saturation is reached. Data saturation has attained widespread acceptance as a methodological principle in qualitative research (Saunders et al., 2018). It is commonly taken to indicate that, on the basis of the data that have been collected or analysed hitherto, further data collection and/or analysis are unnecessary (Guest, Bunce and Johnson, 2006; Saunders et al., 2018). Studies have reported saturation after as few as eight interviews (Cote-Arsenault and Denney-Koelsch, 2011). In a landmark study, Guest, Bunce and Johnson (2006) found that twelve interviews of a homogenous group is all that is needed to reach saturation. Interviews were conducted with parents and healthcare professionals until data saturation was reached. This culminated in a sample of twenty-three parents and twelve healthcare professionals.

A total of twenty-three parents (eighteen mothers and five fathers) participated in the study. Parents' age ranged from 21-47 years (mean-36 years). Eighteen of the parents were married or cohabiting, two parents were separated, two parents were single and one parent was widowed. The number of children in the family ranged from 1-5 (mean-2.43). Eight parents had a degree/diploma; seven parents had completed technical/vocational courses, one parent had a professional qualification, three parents had completed lower secondary education, one had completed upper secondary education, two had completed primary education and one parent had no formal education. Twenty parents were full-time carers for their child and three parents were working for payment/profit. The sample was homogenous in terms of ethnic profile with all parents of Irish ethnic background. Fifteen parents lived in an urban area and eight parents lived in a rural area on the island of Ireland. At the time of the interview the children's ages ranged from 1-16 years (mean-6 years). As previously stated twelve healthcare professionals participated in the study. The healthcare professionals had a variety of roles in CPC service provision including three clinical nurse managers, two social workers, two children's hospice nurse specialists, a paediatrician, a neonatologist, a clinical psychologist, an Outreach nurse and a General Practitioner. The healthcare

professionals provided CPC in a variety of settings including hospital, community, hospice and respite settings on the island of Ireland. The twelve healthcare professionals provided a good representation of disciplines and career stages. All of the healthcare professionals were involved in the assessment and delivery of CPC in excess of five years.

### **4.3 Data-collection**

The data was collected over a twelve-month period. The qualitative data were collected using one-to-one semi-structured interviews with parents and healthcare professionals. Semi-structured interviews were undertaken in order to gather in-depth qualitative data on CPC service provision. They were chosen as they allow participants to describe and explain their perceptions in their own terms. Semi-structured interviews are used when researchers have a list of topics or broad questions that must be covered in an interview (Polit and Beck, 2018). The semi-structured interviews were considered appropriate as there were a number of predetermined topics which needed to be covered, yet the study was exploratory in nature and as such, an element of freedom was required in the interviews (Polit and Beck, 2018). The participants had the freedom to express their views and discuss issues that were important to them which were not always covered in the designed interview guide (Moule, Aveyard and Goodman, 2016). The quantitative data were collected from parents using two standardised measures and a demographic questionnaire developed by the researcher. The quantitative data were collected from parents at the end of their interview. Generally parents' self-completed the standardised measures and the researcher called the questions out in the demographic questionnaire and marked their responses in for them. The selection of the specific standardised measures was informed by a review of the literature. The measures were also chosen on the basis that they could be completed by parents, could be self-administered and had been reported to be psychometrically sound. The standardised measures used were the Decisional Conflict Scale (DCS) and the Measure of Processes of Care-20 Scale (MPOC-20). Details regarding the measurement scales will be discussed in detail following a description of the interview guides.

#### **4.3.1 Interview Guide for parents**

Interview guides were developed for parents to ensure all question areas were addressed (Appendix I). The interview guides included a number of questions, a list of major topics of interest and a list of prompts to facilitate exploration of each area. The questions and topics were constructed from the findings of the literature review and in line with the objectives of the study. Probes as opposed to questions were frequently used. It was important to represent the views of parents and not to lead

them by having specifically designed questions. Discretion was used regarding the order in which the questions were asked. The non-prescriptive nature of the interview guide was viewed as essential as it was vital to concentrate on engaging with parents. The introductory question in the interview guide was designed to encourage them to speak freely about their experiences of caring for their child. The interview guide for parents also contained a number of questions regarding the palliative care services and support that they and their child receive. Additionally the interview guide contained questions regarding decisions that parents made in the past and their involvement in decision-making.

### **4.3.2 Interview Guide for healthcare professionals**

Interview guides were also developed for healthcare professionals to ensure all question areas were addressed (Appendix J). The interview guides included a number of questions, a list of major topics of interest and a list of prompts to facilitate exploration of each area. The questions and topics were constructed from the findings of the literature review and in line with the objectives of the study. The introductory question in the interview guide was designed to elicit information regarding their role within CPC service provision and their experience of providing palliative care. The interview guide for healthcare professionals also contained a number of questions regarding current CPC service provision and the unmet needs for the child and family. These questions were designed to determine the healthcare professionals' perceptions of current CPC services. The healthcare professionals were asked questions regarding barriers and equity in CPC service provision. These questions were designed to determine if there is equal access to services for all families and to highlight any known or possible barriers to services for families. Additionally the interview guide contained questions regarding decision-making in CPC.

### **4.3.3 The Decisional Conflict Scale (DCS)**

Decisional conflict is defined as personal uncertainty about which course of action to take when choice among competing options involves risk, regret or challenge to personal life values (Le Blanc et al., 2009). It is influenced by inadequate knowledge, unclear values, inadequate support and the perception that an ineffective decision has been made. Decisional conflict was measured using the DCS (O' Connor, 1993) (Appendix K). The DCS measures personal perceptions of: a) uncertainty in choosing options; b) modifiable factors contributing to uncertainty such as feeling uninformed, unclear about personal values and unsupported in decision-making; and c) effective decision-making such as feeling the choice is informed, values-based, likely to be implemented and expressing satisfaction with the choice. There are 16 items in the DCS, which measure 5 domains relevant to

decision-making: uncertainty, informed feeling, values clarity, support and effective decision-making. Scale scores range from 0 (no decisional conflict) to 100 (extremely high decisional conflict). Scores lower than 25 are associated with implementing decisions; scores exceeding 37.5 are associated with decision delay or feeling unsure about implementation. The DCS was chosen as it is a well validated and reliable instrument (O' Connor, 1993). Test-retest correlations and Cronbach alpha coefficients exceed 0.78. The construct validity was tested and found that the DCS correlates to related constructs of knowledge, regret and discontinuance. Moreover, it discriminates between known groups: those who make and those who delay decisions (effect size ranges 0.4 to 0.8). Additionally the DCS was found to be a valid and reliable instrument to measure decision-making and decisional conflict within a population of children with life-limiting conditions (Knapp et al., 2009). The DCS is available on line to use without permission once the original reference is cited.

#### **4.3.4 The Measures of Processes of Care-20 - (MPOC-20)**

Family-centred care has been described as a partnership approach to healthcare decision-making (Rosenbaum et al., 1998). The importance of a partnership between parents and healthcare professionals in the support of children with life-limiting conditions is widely acknowledged. The MPOC-20 (Appendix L) measures family-centred care within five domains: Enabling and Partnership; Providing General Information; Providing Specific Information; Coordinated and Comprehensive Care; and Respectful and Supported Care. The measure was originally developed to examine the way in which care is delivered, and the impact this service delivery has on children with disabilities and their families. Each question is answered on a Likert scale from 1 to 7, with 1 representing 'Not at All' and 7 representing 'To a very great extent.' Respondents can also answer 'Not Applicable' to allow discrimination between those to whom an item does not apply, and those who did not receive the behaviour described in the question (King, Rosenbaum and King, 1995). For each of the five domains, a mean score is calculated, resulting in five individual subscale scores. There is no total score because it is thought to be more informative clinically to examine the relationships of the individual scales to other variables. A mean score around 4 indicates that on average, parents report that healthcare professionals 'sometimes' meet parents' needs on that scale. A mean score of 7 (or just slightly less than 7) indicates that needs are being met 'to a great extent'. A mean score of 1 (or very close to 1) denotes that parents' needs on that scale are 'never' (or nearly never met).

The MPOC-20 was chosen as it is a well validated and reliable self-report measure of parents' perceptions of the extent to which the health care services they and their child receive are family-centred (King, King and Rosenbaum, 2004). The MPOC-20 performed well in terms of test-retest

reliability (intra-class correlations from 0.81 to 0.86), and internal consistency (Cronbach's alphas ranging from 0.77 to 0.90). Moreover, with respect to concurrent validity, the MPOC-20 was positively correlated with a measure of parental satisfaction with care ( $r=0.35$  to  $0.72$ ), and negatively correlated with a measure of parental stress associated with the care of an ill child ( $r=-0.18$  to  $-0.48$ ). Social desirability response bias was assessed as well. Rather than gauging socially desirable responses reflecting ideal experiences, responses to the MPOC-20 also appeared to reflect real experiences. The MPOC-20 also demonstrated suitable discriminate validity, in that it was able to discriminate among different parental experiences of caregiving (King, King and Rosenbaum, 2004). The researcher sought and has been granted the permission to use the MPOC-20 scale.

#### **4.3.5 Demographic questionnaire**

The demographic questionnaire was used to gather contextual information regarding the child, the family makeup and other factors which may impact on access and experience of CPC services (Appendix M). Items included in the questionnaire arose from reviewing the literature and consultation with clinical experts. The questionnaire included questions related to the parents, for example age, marital status, number of children in the family, level of education, employment status and ethnic origins. It also included questions regarding where the family resides and services their child receives such as respite care.

#### **4.3.6 The interview process for parents**

The interviews were conducted at a time and location which was suitable for the parents. Twenty out of the twenty-three parents chose to be interviewed in their own home. One parent chose to be interviewed in a quiet room in the children's hospice. One parent chose to be interviewed in a coffee shop whilst one parent chose to be interviewed in a hotel foyer. Prior to commencing the interview, the researcher engaged in general conversation in order to develop a rapport with the parent. The duration of the interviews ranged from 56-133 minutes (average length-82 minutes). Parents frequently attended to the care of the child during the course of the interview, for example, repositioning the child, administering medications and enteral feeds. At the end of the interview and when the standardised measures and demographic questionnaire were complete many of the parents invited the researcher to look at either photographs or alterations they had made to the family home to accommodate the needs of the child that they had referred to in the interview. The researcher thanked the parents for their time following this.

### **4.3.7 The interview process for healthcare professionals**

Similarly the interviews were conducted at a time and location which was suitable for the healthcare professionals. Seven healthcare professionals chose to be interviewed in their work location whilst five chose to be interviewed in a hotel foyer. Prior to commencing the interview, the researcher also engaged in general conversation in order to develop a rapport with the healthcare professional. The duration of the interviews ranged from 27-80 minutes (average length-54minutes). At the end of the interview the researcher thanked the healthcare professionals for their time. The ethical issues relating to the study will be addressed next.

## **4.4 Ethical considerations**

Ethical approval for this study was granted from the Research Ethics Committee in Dublin City University. Ethical approval was also granted from each of the five aforementioned organisations. These included: the Children's University Hospital in Temple Street, the University Hospital Complex in Limerick, Letterkenny University Hospital and Saint John of Gods. Ethical approval was also granted from the Office of Research Ethics Northern Ireland (ORECNI).

### **4.4.1 Informed Consent**

All the participants were fully informed about the study and were free to make a choice regarding participation. Before making a decision about whether or not to participate all potential participants were issued with a Letter of invitation and Information which provided comprehensive details of the study. The Letter of invitation included a contact telephone number and email address for the researcher. The potential participants were invited to contact the researcher if they required any additional information or sought to discuss any aspect of the study before making a decision regarding participation. The participants were given time to consider the information and given the opportunity to ask any questions about the study prior to seeking consent. All participants were informed of their right to participate or not in the study. Parents were made aware that the service they or their child received would not be affected in any way by their decision regarding participation. This was seen as particularly imperative as individuals may feel that their care could be adversely affected if they did not agree to be research participants (Grove and Gray, 2018). All of the participants were informed that the interviews would be audio recorded with their permission. The Consent forms (Appendix D and Appendix H) were prepared in a manner to convey as much information as possible in clear and concise language. Additionally the consent forms included a component relating to the recording of data to ensure that participants agreed to their interview

being recorded. Two copies of the Consent were taken one for the participant and the other retained by the research team.

#### **4.4.2 Confidentiality**

Confidentiality was maintained through the following procedures. The recordings of the interview were transcribed and all identifying information was removed such as names, diagnosis and named services. Written copies of transcripts were stored in a locked cabinet in the researcher's home, where the researcher is the only person with access. Digital recordings of the interviews were stored on the researcher's laptop which is password protected to ensure confidentiality of all electronic records. The records of signed consent were also stored in a locked cabinet away from the written transcripts. All data returned in the demographic questionnaires and standardised measures were treated as confidential. 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. All data records will be retained in locked storage as previously described for this period.

Green and Thorogood (2018) state that researchers cannot offer complete confidentiality in research settings as there are situations in which there may be an obligation to break this. Additionally, assurances of anonymity and confidentiality need to be realistic (Green and Thorogood, 2018). The limits of confidentiality were explained to participants at the outset and were also outlined in the information sheet and consent form. The researcher informed participants of the requirement to disclose information if matters relating to the safety of a child were raised during the interview. Furthermore, in highly specialised areas the identity of participants can sometimes be logically deduced. Parents were advised that there was a slight possibility that given the relatively small population of children with life-limiting conditions, that some accounts would be recognisable to familiar others even when anonymised. Although the researcher made every effort to remove such opportunities for others to deduce the participants' identity from aggregated data, it was important to preserve enough identity to give the reader sufficient context to understand the findings. Participants were also advised of this limitation in the information sheet and consent form.

#### **4.4.3 Subject sensitivity**

Hennink, Hutter and Bailey (2011) suggest that the process of interviewing, particularly on sensitive issues, can evoke emotional responses from participants that researchers have to be prepared for with empathy and professional support. Occasionally participants may become unexpectedly emotional and researchers need to show understanding, and consider terminating the interview if it

is causing distress. The researcher was cognisant of the sensitive nature of the research. The participants were not pressurised in any way to talk about experiences, which appeared to upset them. The researcher developed a specific protocol that could be used if a participant became upset during the interview. The steps in the protocol included stopping the interview if a participant showed any signs of distress. The researcher would ask the participant whether he/she would like to terminate the interview or alternatively to take a break. The researcher would remain with the participant and offer immediate support for as long as required. The interview would only be recommenced if the participant indicated that he/she would like it to be recommenced. Support structures were also in place for parents and healthcare professionals if additional support was required. The researcher who conducted the interviews had experience of interviewing and supporting parents and families. The researcher was experienced in dealing with emotional issues that can arise during interviews. Additionally the researcher undertook specific interview training. Where interviewees became upset during the interviews (as happened in a number of instances) they were offered the option of discontinuing the interviewing or taking a break. All declined and wished to proceed. After a difficult interview, the researcher remained with, and provided support to, the interviewee as required. No participant required referral to additional services. Researcher health and safety was also considered. Researchers may be at risk of experiencing emotional exhaustion, especially if provided with insufficient training or support (Dickson-Swift et al., 2009; Petty, 2017). The researcher was conscious that interviewing can be intense and at times distressing as parents recount painful aspects of their lives. Scheduling of the interviews was considered important to allow time between interviews. Support structures were also in place to allow the researcher to debrief. The researcher used the support structures in place, particularly at the beginning of data-collection.

#### **4.5 Data analysis and integration**

In terms of the qualitative data thematic analysis was used in the analysis of the interviews. Braun and Clarke's (2006) six-phase framework for purposeful, systematic and rigorous thematic analysis was used to help guide the analysis of the qualitative data. Thematic analysis was chosen as it offers an accessible and theoretically flexible approach which has the potential to provide a rich description of the data (Braun and Clarke, 2006). According to Elo and Kyngäs (2008), thematic analysis is commonly used in nursing studies, but there has been a limited publication of the actual analysis process and generally they only provide a brief description. However, Braun and Clarke (2006) have provided comprehensive guidelines on the process of applying thematic analysis. These guidelines were used and are outlined in Table 7 below:

**Table 7: Braun and Clarke (2006) six phases of thematic analysis**

Phase	Description of the process
1. Familiarising yourself with your data:	This involved transcribing the data, reading and re-reading the interview transcripts, noting down initial ideas
2. Generating initial ideas:	Coding interesting features of the data in a systematic fashion across the entire data set, collating data relevant to each code.
3. Searching for themes:	Collating codes into potential themes, gathering all data relevant to each potential theme.
4. Reviewing themes:	Checking if the themes work in relation to the coded extracts (Level 1) and the entire data set (Level 2), generating a thematic map of the analysis.
5. Defining and naming themes:	Ongoing analysis to refine the specifics of each theme, and the overall story the analysis tells, generating clear definitions and names for each theme.
6. Producing the report:	The final opportunity for analysis. This involved selection of vivid, compelling extract examples, final analysis of selected extracts, relating back of the analysis to the research question and literature, producing a scholarly report of the analysis.

In the process of thematic analysis there are two ways to identify themes or patterns. Firstly, an inductive approach means that the themes identified are strongly linked to the data. Secondly, a theoretical deductive approach is when the structure of the analysis is influenced by the researcher's theoretical interest in the area or topic (Braun and Clarke, 2006). An inductive approach was used in this study. First, the researcher became familiar with the data through transcription of audio recorded interviews and reading/re-reading the transcripts (phase one). Phase two included the generation of initial codes, whereby the researcher systematically coded the data line-by-line in each transcript across the entire data set whilst collating data pertinent to each created code. At this stage a coding framework was generated. Subsequently, the transcripts and audio recordings were imported into NVivo (Version 10) computer software, organisational software that facilitates the storage, retrieval and management of large quantities of qualitative data. Third, all codes were collated and then synthesised into themes and subthemes. Fourth, all codes, themes and subthemes were reviewed by members of the research team with interpretations validated and substantiated through discussion and repeated referral to the coded extracts and original dataset. Fifth, definitions for the themes and subthemes were developed. The entire process culminated in

the production of a written report of the analysis (phase six), whereby the researcher selected pertinent and compelling extract examples to present in order to address the research questions, relating findings back to previous literature (see Chapters 5, 6, 7 and 8). In terms of the quantitative data which included data from the demographic questionnaire and standardised measures SPSS was used for the management and to aid statistical analysis of data. Recognising the exploratory nature of this research and the small sample size, quantitative analysis was limited to descriptive analyses, cross section analyses and correlational analyses (see Chapter 5). A full outline of the analyses conducted is given in the said chapter. In the spirit of a mixed methods analysis, qualitative and quantitative data were integrated.

Integration is an intrinsic element of mixed methods research (Creswell, 2017; Johnson, Onwuegbuzie and Turner, 2007; Moran-Ellis et al., 2006; Tashakkori and Teddlie, 2010). Numerous guides and frameworks have been provided for the integration of findings uncovered through mixed methods research (O' Cathain, Murphy and Nicholl, 2010; Tashakkori and Teddlie, 2010). O' Cathain, Murphy and Nicholl (2010) outline three techniques for the integration of findings uncovered through mixed methods research, these are; 1) triangulation protocol, 2) following a thread, and 3) the use of a mixed methods matrix. Triangulation protocol was deemed the most appropriate form of integration for the present study as it allows for the interpretation of qualitative and quantitative findings following the separate analysis of both data sets. The data elucidated from the present study will be analysed and discussed separately. Subsequently, the findings will be integratively discussed in order to provide a greater understanding of the topic under investigation; the experience of caring for children with NMLLCs including, accessing services and decision-making surrounding the child's care. The integrative discussion of the findings will be guided by the triangulation protocol described by Farmer et al. (2006). This process of integration of the findings allows for several possible outcomes convergence (where data sets agree or are consistent); complementarity (where data sets offer complementary information on the same issue and therefore enhance knowledge and/or understanding of the phenomenon under investigation); silence (where one data set uncovers an issue whereas it appears silent in the other data set); and discrepancy or dissonance (where the data sets have conflicting findings) (Farmer et al., 2006; O' Cathain, Murphy and Nicholl, 2010).

## **4.6 Concluding comments**

To conclude, in order to address the research aims and objectives, a concurrent embedded mixed methods design, underpinned by the philosophical paradigm of pragmatism, is implemented.

Collectively, the qualitative and quantitative data will enable an enriched understanding of the experience of caring for children with NMLLCs including, accessing services and decision-making surrounding the child's care from the perspective of parents and healthcare professionals; a topic that was previously underexplored and under-researched in CPC literature. The qualitative and quantitative research findings from the parents will be presented in the subsequent chapter.

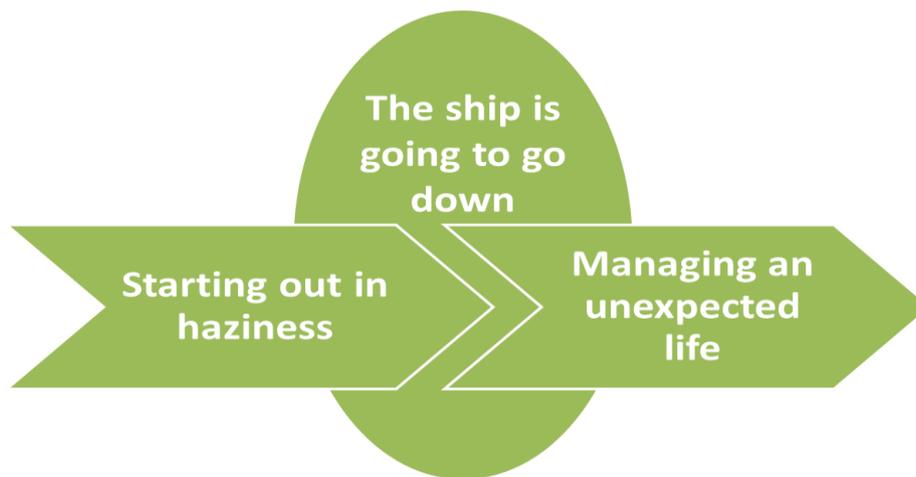
## **Chapter 5: Presentation of Findings: Parents**

### **5.0 Introduction**

This chapter will present the qualitative and quantitative research findings from the parents in this study. Thus, this chapter will present the findings from the data analysis carried out on the twenty-three parent interviews. The quantitative findings from the analyses of the two standardised measures, namely the MPOC-20 and the DCS and aspects of the demographic questionnaire such as the data related to respite care will also be presented. As indicated in the previous chapter, Braun and Clarke's (2006) six-phase framework for purposeful, systematic and rigorous thematic analysis was used to help guide the analysis of the qualitative data. Thematic analysis is one of the most common forms of data analysis used in qualitative research (Guest, MacQueen and Namey, 2012). It offers an accessible and theoretically flexible approach to analysing qualitative data. A detailed outline of the processes through which thematic analysis was undertaken is presented in the previous chapter. Quantitative analysis centred on descriptive analyses, cross section analyses and correlational analyses. In this chapter the qualitative findings will be presented first. Following this the quantitative findings will be presented. The qualitative and quantitative research findings from the parents will be integrated in the subsequent chapter.

### **5.1 Overview of qualitative findings**

Data analysis was inductive in nature i.e. the themes emerged from the data. This resulted in three dominant themes and eight subthemes emerging, charting the parents' journeys and unique experiences of caring for a child with a NMLLC, including the particular issue of access to services and decision-making surrounding their child's care. The three dominant themes are diagrammatically represented in Figure 5. The relationship between the three dominant themes are then discussed. Following this each theme is systematically unpacked. Explicit discussion of their relevance or how they answer the research objectives will be undertaken subsequently.



**Figure 5: Diagrammatic representation of themes: parents**

## **5.2 Relationship between the themes**

During ongoing thematic analysis of the parents' interviews a chronology became apparent across Theme 1 'Starting out in haziness' and Theme 2 'Managing an unexpected life'. Each of these themes marked a distinct period of time with each theme/period of time containing its own specific issues in relation to caring for a child with a NMLLC, including the particular issue of access to services and decision-making surrounding their child's care. Theme 3 'The ship is going to go down' contained specific issues which were present and pervasive throughout the parent stories and each of the two themes. The relationship between the themes is also illustrated in Figure 5.

## **5.3 Starting out in haziness**

This analytically derived theme arose from each parent account and was marked by the plethora of overwhelming and palpable emotions inherent in the stories recounted. The theme 'Starting out in haziness' represented the time period from diagnosis or recognition of a life-limiting condition and marked the beginning of an unknown journey filled with emotional turmoil. As parents were 'Starting out in haziness' they became aware that a number of actions were now necessitated by them as parents. The awareness of these actions and the action itself marked this time period and became the subthemes that made up the overarching theme, those being 'Entering a whole new world', 'Acquiring an invisible L plate' and 'Navigating the unknown territory'. Please see Figure 6.



**Figure 6: Diagrammatic representation of Theme 1**

For some parents the diagnosis came early either antenatally or shortly after the birth of their baby. The unexpectedness and speed of the diagnosis often left parents feeling ill-prepared to deal with the situation:

*Yes, at my first scan I found out there was a problems. You think going in for your first scan you are going to get great news, you don't think you are going to be told that there is no hope for your child. [P8].*

Other parents faced a long and arduous search for a diagnosis which sometimes took months or years. Receiving a named diagnosis was very important for these parents: *“we fought for a diagnosis to find out where we were going and what we were doing and would things change and what other people’s experiences were.” [P2].* Often parents showed a determination to arrive at a diagnosis that made sense of their situation. Parents also felt that a diagnosis would provide a degree of clarity or direction as they were starting out on their unknown journey. Sometimes, however the specific diagnosis was never found. This lack of a named condition often left parents feeling in limbo:

*She carried out a huge array of tests, everything you could think of. Everything came back negative. She said that there was no doubt in her mind that there was something seriously wrong with him. She didn't have any idea what it was, that all the obvious tests had come back negative, and sometimes it can take time for things to present. So, it was a waiting game. [P18].*

Parents who did finally receive a named diagnosis realised that it did not offer a solution to the many challenges that they faced.

Whilst parents' stories were all individual and the presentation of their child's condition different what was clear was the diagnosis or recognition of a life-limiting condition was life-altering. It shattered dreams, transformed lives and propelled most into recognising that their parenting experience would be different:

*I had to bury my dreams of what he was going to be. We'd got married, had (sibling's name) two years later after that had (child's name), so I had a girl and a boy, so that's every person's dream really, and the boy was going to play rugby and was going to be everything that I wasn't, and next thing I have a child you were going to be pushing in a wheelchair for the rest of your days. So you have to bury that dream, you have to come to terms with that, and you have to mourn that. [P2].*

Additionally, it appeared that this was the first time in their lives that they had encountered such an overwhelming inability to alter or influence events.

All parents experienced profound emotional distress at this time. This emotional distress triggered the haziness:

*I know I cried a lot, but I remember that day to me it was one of the worst days. I remember just being so hysterical and going down to the car because I knew I couldn't go up to the ward because I was hysterical. It was the first time and last time I ever thought this thought, but I did think, will I just get into the car and drive into a wall? [P4].*

Parents described how they were utterly shocked at this time. As a result they could not comprehend the information that was imparted to them:

*I couldn't understand what was going on. I couldn't. They were talking about PDA's and ....and all this stuff that I just didn't understand. I just couldn't. I had to step back because I couldn't understand what they were talking about. [P3].*

Grasping the immediate and long-term implications of the diagnosis was a complex process for all the parents. Parents spoke of having uncertainty; of not knowing the extent of what having this condition meant for them and their child and of feeling that they had no real conceptualisation of what the future may bring: "they said her head was small. They kept telling me her head was small. I didn't understand what they meant. I thought her head was small because she was premature." [P11]. Parents reported that they felt powerless at this time.

Parents described the initial months following diagnosis or recognition of a life-limiting condition as life-altering and all-consuming as they were starting out in haziness. The following quote echoed sentiments expressed by a number of parents: *“Oh my God! It was a nightmare. I was nearly going out of my mind.”*[P1]. A persistent state of fatigue was described by all parents. Parents reported the unremitting nature of caring for their child who often required care throughout the day and night:

*Because we were feeding him on demand, he was only taking small feeds, but the feeds were very often. So if it was 1 o'clock he needed a feed, that was fine and if it was half-2 he needed a feed, that was fine, and that's the way we worked with him. [P14].*

This sentiment was mirrored in the following quote:

*Oh, we were exhausted and being able to do nothing else but always somebody holding him. You couldn't even put him down to have dinner. It was like one person holding him, the other person having dinner. [P19].*

Parents described how meeting their own basic needs became secondary due to the care demands of the child.

Low mood was something that was a particular issue shortly after diagnosis in the haziness that marked their lives. A number of mothers and fathers reported that they were prescribed antidepressant medication in the months following diagnosis. Parents grieved the loss of a healthy child at this time. Furthermore, they also grieved the loss of their dreams and aspirations for their child:

*I suppose it's like a grief, you go through all the different stages, you go through the sadness, the loss of the child that you were expecting to have, you know, the grief of having to look at a baby not developing. It would break your heart. [P10].*

Despite a backdrop of overwhelming emotion parents recognised that life as they knew it was permanently altered: *“we thought our world was going to fall apart.”*[P15]. Parents were aware that they were ‘Entering a whole new world’.

### **5.3.1 Entering a whole new world**

As parents were ‘Starting out in haziness’ they were aware that they were simultaneously ‘Entering a whole new world’ one characterised by hospitals, appointments, medical investigations and equipment. It was also a world characterised by decision-making that they as parents imminently became part of. Parents recounted how they themselves did not actively choose to be part of this world: *“I entered a wee world that I didn't know existed beforehand. I never ever wanted to be part*

of. *And you are part of it.*”[P6]. Moreover, entering the new world was a journey into a world marked with unfamiliarity and a degree of intimidation:

*Because it's a different world, you know. When you go into one of those rooms, it's a live-or-die room. It's an alien world because you've never been in to an intensive care unit, and you don't understand what it's about.* [P15].

Their personal world had become completely uncertain and unpredictable: *“everything's all new to you.”*[P11]. Additionally, their hopes, dreams and plans no longer seemed to pertain as the world that they knew was altered irrevocably.

On entering this new world parents entered a world of decision-making that they had never known. Parents faced numerous challenging decisions, including decisions regarding ventilation, surgery and invasive treatments. Parents revealed decisions that created incredible stress for them and their families:

*We were told in the meeting about the operation that if she didn't have the operation she would die but there was a possibility that she could die in surgery as well. So it was a case of we just had to take that lifeline and hope for the best.* [P12].

Within a single conversation, parents were often given the diagnosis, the possible prognosis and then presented with a complex array of decisions. All parents wanted to give their child every chance of survival at this time: *“he was on life support and lying there, and we had told the doctors, you know, no matter where you have to take him, just try and keep him safe.”*[P14]. Parents were often faced with the immense, heart-breaking responsibility to withdraw life-sustaining treatments for their infant or child at the time of diagnosis. Additionally, parents were often required to make these decisions in a dynamic and time-sensitive manner.

A number of parents regretted the decisions that they made at the time of diagnosis as they proceeded on their care journey. Often these decisions were made in moments of profound emotional turmoil:

*We decided not to shunt him, and we brought him home. I don't know was that the best decision? In hindsight, now, I regret that we didn't shunt him earlier, because his head got bigger and bigger and bigger, and we didn't shunt him until he was seven weeks old, and his head, now, at that stage, was massive, so it was, which in hindsight I regret. I wish we had.* [P19].

Sometimes the outcome of certain decisions could not be predicted. This is described in the following scenario: *“so we made a decision then that night that we were going to take him off the*

ventilator the next day, because the longer we left it, the worse it was going to get.”[P10]. The baby however, survived following the withdrawal of ventilation:

*We decided to have him christened the next day and we got his godparents together and they came into the hospital. We got all our family together to say goodbye, because they gave us an indication that he wouldn't survive. So they all came in and said their goodbyes and we had him christened at about 5 o'clock and everybody left, and then at 7 o'clock we took him off the ventilator and he started breathing by himself. [P10].*

The haziness undoubtedly had a significant impact on the parent's ability to make decisions at the time of diagnosis or recognition of a life-limiting condition. In addition, parents also had to acquire a new range of knowledge and skills in the new world that they had now inadvertently become part of.

### **5.3.2 Acquiring an invisible L plate**

As parents were 'Starting out in haziness' they had to acquire a new range of knowledge and skills as they learnt to parent in a new and unexpected way. Learning to care for their child was often perceived as an experiential undertaking:

*As I always say to people, there's an invisible L plate. You can't see it, but Mummy and Daddy have an L plate. We're only learning...we haven't been actually trained to do this... we're teaching ourselves. We haven't got it wrong yet but there's no saying we won't get it wrong, but we'll not deliberately get it wrong. [P15].*

The knowledge and skills were distinct from the knowledge and skills required with becoming a parent.

Parents experienced an overwhelming sense of fear in the early days of learning to care for their child. This fear generally stemmed from their perceived lack of knowledge, the fragile nature of the child and the health status of the child: “we were worried sick about him, because he was very, very small.”[P19]. Parents discussed specific fears for example, fears of dislodging endotracheal tubes and oxygen tubing and of those fears, then contributing to reservations about touching or holding their child: “I just got to hold him, it was a mixture of relief, love, and worry that, you know, am I going to knock his tube out here? What if he stops breathing on me?” [P14]. Parents described being overwhelmed, particularly in the early stages of coming home from hospital with their child: “when we first came home, I didn't go to the toilet. I was terrified.”[P3]. This sentiment was reiterated in the following excerpt:

*and we got him home the day before Mother's Day which was quite nerve-racking and in fact the first night we didn't sleep a wink because we stayed up and kept checking him in the cot, worrying is he breathing, is he alright? [P14].*

Parents learned ways to respond to their fears, for example, holding their infant/child despite the worry of dislodging tubes, constantly checking the infant/child or making a conscious decision not to sleep. However, worry, the need to remain constantly alert and sleep deprivation exacerbated the haziness.

Providing care at home necessitated that all parents learned skills akin to those usually performed professionally by nurses. In most cases the level of specialised care required by the child precluded obtaining help from family or friends. Parents described the challenges of providing technical care. Parents recalled having concerns during the period in which they were being trained for and/or took on responsibility for a procedure. Many parents reported that they were extremely anxious at this time:

*I never learnt to do it like. I just did it once and it really upset him and I caused him problems. It didn't work out right. I couldn't do it again. Maybe now I would be able to do it. I couldn't do it at the time. [P1].*

Parents reported having concerns about the safety implications of undertaking certain procedures and as a result, they often needed reassurance from healthcare professionals:

*They showed me the PEG how to clean it one day and I nearly got weak. The nurse was lovely. She was really nice. She showed me how to do it about ten times. I said, 'what if it comes out?' she said, 'it won't.' She was really good, like, in fairness to her. [P1].*

The responsibility for a procedure that was potentially life-threatening if mishandled was daunting. However support from healthcare professionals made 'Acquiring an invisible L plate' easier.

It was clear that parents were often overwhelmed and ill-prepared with the experience of learning to care for their child at the time of diagnosis or recognition of a life-limiting condition. However support from healthcare professionals and social support made the experience easier. Parents were also faced with the issue of accessing services and support for their child at this time.

Communication with healthcare professionals played a significant role in how parents began the process of 'Navigating the unknown territory'.

### 5.3.3 Navigating the unknown territory

Navigating the unknown territory refers to the process that the parents were required to undertake in order to access services and support in caring for their child. It was clear from the parents' stories that the experience of caring for a child with a NMLLC was also overwhelming in terms of finding and accessing services and support.

Parents' ability to navigate the unknown territory was strengthened through the provision of information in a sensitive and timely manner. However, for many parents, the communication process at the time of diagnosis was fraught with difficulty. Insensitive use of language impacted significantly on parents as they were starting out in haziness:

*They basically said to me that my son had been born a vegetable, and that's the word that they used. They told me that he was going to have no quality... if he were to survive; he would have absolutely no quality of life whatsoever. [P10].*

Similarly this is evident in the following excerpt:

*He came to us and said, basically, your child will never walk. He will never sit up. He will be a vegetable. He will be on a sofa, on a ventilator 24hrs a day for the rest of his days. That is the condition that is the future. [P16].*

While learning about the diagnosis, the parents were also processing the emotional impact of this unexpected, often life-changing situation.

Initial information was often given at a time of intense emotional distress. Many parents described how they were given heart breaking information without any follow-up support. For some parents, the stress of the diagnosis was heightened by their perceived lack of professional support:

*And what we were actually told, you've got a diagnosis of (name of condition). And the exact words were, well, I suppose you'll have to go home now and make the best of the time you have. And for us, I think that was, in my mind, far more devastating than being told (child's name) had (name of condition). It was the fact that because when it's (name of condition), there's nothing because there's no treatment and there's no cure. It was the nothingness of it. [P18].*

Furthermore, many mothers in particular described scenarios where they were given distressing information when they were alone and unsupported:

*They brought me up to a room to have a wee chat with me. Again, a lot of it is hazy, but I feel like I'm angry that they didn't wait until (husband's name) was there and bring both of us up. I would be very angry with them. [P4].*

This feeling was also presented in the following excerpt:

*So, the lovely neonatologist pointed at me and said, is he your child? And I said, no, she's my child. And he said, well, there's something wrong with her. We've done an ultrasound. There's fluid on her brain. It's very serious and she'll be off to (name of Children's hospital) in the morning. So, of course, me, 24 hours after giving birth, no husband with me, and no idea what was going on? [P21].*

Perceived lack of support left parents feeling overwhelmed as they were starting out in haziness.

Many of the concerns and information needs expressed by parents involved a need for more information. Several parents commented that healthcare professionals lacked knowledge regarding their child's condition at the time of diagnosis:

*He just said, your son has (name of syndrome) and he just said there's a leaflet. I don't know anything about it. You'll have to wait and talk to another consultant from the metabolic unit, and just sent me on my way. The leaflet he gave me was more so about another syndrome, and there was only one paragraph about (name of syndrome), which didn't give an awful lot of information. [P22].*

Parents also described how they were not given enough practical information:

*I didn't realise there was parents' accommodation either. Nobody told me about that. At the time I didn't know there was such a thing as parents' accommodation. I never asked. I didn't know. I was driving home at night, it would have been better if I could stay. I might have left the hospital sometimes at twelve or one o'clock.... going home, expressing, going to bed and getting back up again. [P4].*

Parents also perceived reluctance among healthcare professionals to share information with them:

*The neonatal doctor was very, like... he would never answer my questions. He stepped out of the door like this as I was asking questions. He was going backwards out the door. He never wanted to answer me. He'd come in and go, alright, so today we've got the Newcastle Work up and the bloods, whatever, and I hadn't a clue what all this meant. He'd be backing out the door when I asked questions. He'd be gone. [P4].*

Parents were frustrated with the lack of information. All parents talked about how they sought out further information soon after receiving their child's diagnosis. In some cases they talked about needing to do so partly because healthcare professionals had given them insufficient information, but for all of them it seemed to be a useful coping strategy, a feeling they were taking charge as they were starting out in haziness.

Hence 'Starting out in haziness' represented an emotionally challenging time period for all parents as they began their unknown journey. Three elements were central during this time period those being 'Entering a whole new world', 'Acquiring an invisible L plate' and 'Navigating the unknown territory'. Eventually parents transitioned from this time period and began the process of 'Managing an unexpected life'.

#### 5.4 Managing an unexpected life

This analytically derived theme captures the time period after the haziness started to settle in the component of the parents' stories and where they attempted to respond to the resultant emotional and practical impact of their child's condition on their life and that of their family. 'Managing an unexpected life' involved three specific areas which arose as subthemes those being 'Striving for normality', 'Becoming the expert in their child's care' and 'Fighting for your child'. See Figure 7.



**Figure 7: Diagrammatic representation of Theme 2**

Parents described that they came to realise that they had no choice other than to move forward and make some attempt at managing their new and unexpected life. They needed to move forward and continue on in life, even though they now faced a different life journey, one that was not part of their original plan. However, they could only move forward and begin to manage when the intense emotions experienced at the time of diagnosis or recognition that their child had a life-limiting condition dissipated:

*I was so so upset when it happened first and I was crying, and crying and crying. But crying wasn't going to change anything. I had to get up and get on with it and do the best that I could. [P17].*

What was clear was that parental distress had not disappeared completely, but rather that those emotions were not so intense or overwhelming as they were at the time of diagnosis or recognition that their child had a life-limiting condition. Moreover, parents highlighted that accepting and managing this new life was incredibly daunting and difficult. A few aspects apparent in parent narratives made the initial management challenging.

Firstly, inherent in coming to terms with their situation was a prolonged process of questioning:

*I did do the whole 'why me' and the guilt, and did I do something wrong? And is this karma? And I didn't smoke, I didn't drink, but maybe I should have done something differently, and all that. I did all that, but then somewhere along the way you've got to just pick yourself up and just crack on, because otherwise it will eat you away. [P3].*

Parents suggested that this process of questioning was exhausting and almost immobilised them and prevented them from devising the strategies now required for managing their new and unexpected life. Secondly, parents reflected on how they had initially made a constant comparison between the child with the life-limiting condition and the healthy child that they had hoped and expected to have. Incidentally, parents made this comparison, irrespective of the timing of the diagnosis. Again, this process was exhausting for parents and held them back from developing the new ways necessary to manage their life and that of their family: *"you'd wear yourself down if you thought too much about what should have been or would have been."*[P17]. However, in time parents began to replace their hopes and expectations with the reality of their child's diagnosis and the need to manage the life they now faced:

*Yes. Like some days it absolutely breaks your heart, but other days you are so elated with the smile that he gives you. You would have taken that for granted previously if the baby was okay, do you know what I mean? [P10].*

The ability to be able to reclaim the simple things in life, such as a smile was vital for parents as they began to manage.

Undeniably, there were times where parents were reminded of what they had lost as they watched others move through life's milestones such as starting school and birthdays:

*He should be starting school in September. It's very hard when occasions like that come around, little milestones and you wonder what would he be like running into school and what*

*sort of child would he be? Would he be as quiet and placid and good as he is now? Because he's such a ...he's a little pet. Or would he be more boisterous? It's something you'll never know. You do think about it a lot. [P17].*

Parents described grieving anew at these times. However, having a practical focus appeared to assist parents in coming to terms with the situation. Parents recalled how they had to move forward and care for their child:

*I was utterly devastated. But you just have to get on with it. There's nothing else for it. It's not her fault. That wee one was lying in your arms and the two of us just wanted to do whatever we could, for however long we had her. [P6].*

Parents appeared to manage this new and unexpected life out of a combination of sense of duty as well as sheer love and devotion. 'Managing' however, was never a fully accomplished skill for parents. Changes and deterioration in the child's condition altered life again, posed multiple new difficulties and frequently reignited a grief reaction for parents:

*She was feeding just like a normal baby. It was the only normal thing that we could do, so when it was taken away it was quite hard. I suppose we thought she'd get back feeding, but she didn't. [P21].*

The sense of loss and consequent grieving occasioned by one such change is palpable in the comment above.

Parents over time appeared to realise that managing this new and unexpected life was a continuous and always unfinished business. This is evidenced in the following quote when a father was informed his child was deaf: "we were devastated, because we didn't think there was anything else wrong with him." [P15]. Along with such realisation parents appeared to be more resilient as time progressed and instigated new practical approaches almost immediately:

*Just knowing that she couldn't see was really, really... it was so upsetting for us. But we had to realise that she didn't know the world that we see. We were kind of mourning for what we can see and what we can do, but she has never been able to see, so for her it's just about making everything nice and comfortable, and lots of tactile stuff and that for her. [P21].*

This sentiment was also illustrated in the following quote:

*When he got diagnosed as being legally blind there a couple of weeks ago, I fell to pieces. And then I picked myself up and I thought well, it's just another... we've just got to find a way to cope with it. [P14].*

Managing also involved making choices and decisions about their child's care. The choices were not easy for parents and involved decisions around, for example, giving up work and future pregnancies.

Decisions were made in the context of the family unit as a whole. Parents, however, acknowledged that normal family functioning was profoundly altered and new ways of managing had to be found: *"Life totally changed."*[P20]. Parents nonetheless shared a common aspiration for a 'normal' life. Parents viewed normalisation as a positive response to managing their child's condition. This is illustrated in the following quote: *"in my eyes she's normal."*[P11]. This feeling was emulated in the following comment: *"he was a baby to me."*[P16]. Creating and achieving normality was not easy for parents. A number of parents eventually relinquished the goal of achieving normality: *"you just want the normal, but you can't have the normal."*[P18]. However, in an attempt to manage parents strove to maintain normal routines and create a 'new normal'. Thus 'Striving for normality' arose as a subtheme as it was a key component to 'Managing an unexpected life'.

#### **5.4.1 Striving for normality**

A core component of parents' management strategies within their new life was undoubtedly their determination regarding their shared common aspiration for normality:

*I wanted as much normality for the length of time that I had with her, like that.... friends calling to see her and family calling to see her, just to have some bit of memories, not just memories of the hospital. [P8].*

Achieving some degree of 'normality' was very important and indeed something to celebrate:

*I can remember his sister bought a pram for us, when she heard that we were coming home and I couldn't wait to walk down the town like any other new mummy would, showing off their baby. [P8].*

On the other hand, parents grieved the loss of normality as they continued on their care journey coming to realise that life would never be the same again. With this awareness came the determination and focused effort to create a 'new normal' necessitated by their child's life-limited condition. The diagnosis or recognition of a life-limiting condition impacted on every aspect of their lives. Therefore the need to regain control, equilibrium and stability for the child, themselves and family were paramount in parent accounts.

Family life was completely disrupted as life needed to revolve around caring for the child with the life-limiting condition:

*From when we went home, for the first six months, it was just a litany of hospital appointments and meeting up with Occupational therapists and speech therapists and physios and we went to the dental school. We got her brain scanned. You name it, every department we were in or generally, I was in because I was the one that was off. [P6].*

Navigating appointments took up a great deal of time in addition to the daily care giving duties. However, the reality was that parents had to meet the care needs of the whole family. Parents had to learn to juggle with the many competing demands of family life together with caring for their child.

Disruption to normal work life for parents was inevitable, with many reducing their hours or leaving altogether in order to manage to care for their child:

*I haven't gone back to work since she was born for a variety of reasons. She was just so sick. The first two years she was in and out of hospital. She had seven operations, five on her hips and one on her nose and then the gastrostomy tube as well. [P6].*

The complexity of the child's condition also made returning to work impossible as no suitable childcare could be arranged. Parents described how work had previously been an important part of their lives and identity. Parents missed the social opportunities that work outside the home presented. Thus the new normal created by parents was absent of either partially or completely of the social opportunities they once experienced through their work.

In the continuous strive for a 'new normal' relationships between parents changed. Parents reported that they had very little time and energy for each other due to the relentless care required by the child:

*I think it's a massive change to a relationship, and I think you do become different people. And you go through good times and bad. It will never be the same again. There's very little time to be together. [P21].*

Parents had to learn to adjust to the impact on their relationship, for some regrettably this did mean separation: "It just kind of took its toll it was a lot on a relationship." [P8]. Other parents stated that they rarely, or never, spent social time with their partner. For many parents this time was just not available, for others the complexity of the child's condition made getting out together impossible as no suitable childcare could be arranged:

*Socially it's very, very, very hard to get out. And like you couldn't just pay the girl around the corner to come in and mind him because it's just not an option. We tend to go out separately if we are going out. [P10].*

Parents regularly became housebound as a result of the intensity of care in the home, and this alongside the absence of socialisation through work led to parental feeling of isolation and being 'cut off' from their once important social world.

Furthermore, parents outlined how the care demands required by the child frequently led to parental guilt regarding the impact on siblings:

*Well, they have missed out on a lot, like. They don't always understand. I feel maybe they're a bit neglected, like, because so much time is with (child's name), like, you know? We do our best for them of course. (Sibling's name) is involved in football so either one of us will bring him.*  
[P1].

In the 'strive for normality' adaptation was required, whereby one parent frequently responded to the needs of the child with the life-limiting condition whilst the other met the siblings' needs.

Emotional and behavioural changes were, however common in siblings: *"he's only seven, and it did have a big impact on him. Worry, anxiety, just... God love him. It did affect him quite badly."* [P20].

In an effort to create normality parents reported that they needed to make a concerted effort to spend time with their other children and as a family:

*So they love when (child's name) is at home with the family. They don't really like taking him out of that. But they understand that when he goes into respite Mum and Dad have time to... you know, go down to the park and to go to the movies. We used to kind of treat them when he went into respite, but we don't do that anymore, we just say to them, look, this is time for Mum and Dad to read you stories and we'll go and watch your football match. So they realise, to a certain extent, that respite is needed because he is hard to mind.* [P10].

Thus, many parents reported that the real value of respite was the opportunity it afforded to spend time with their other children.

The isolation often experienced in the 'unexpected world' that parents found themselves in was exacerbated as support from friends often disappeared after the initial period of diagnosis or recognition of a life-limiting condition:

*I think the longer you're on this journey, I think the more people you lose along the way from your previous life, if you want to call it that. It's almost like, you know when they say before Christ and after Christ, in ours it's before (child's name) and after (child's name).* [P18].

Parents often excused this perceived abandonment of their friends as a difficulty in truly appreciating the complexities of caring for their child and juggling the competing demands of their new life:

*They didn't take on board how much my life had changed, and how I suddenly wasn't around for coffee every week, or to go out at the drop of a hat; that I had to plan and organise if I was going to go out.* [P17].

Parents frequently lamented the loss of spontaneity and spoke about the effort required to arrange an outing.

Parents often turned to other parents in situations similar to their own as a life-sustaining source of support:

*Because I know I can ring one of the girls, no matter what part of the country they're in, and just go, do you know what? Today is really (curse). And they'll be like, oh, yes, yes. Oh hold on a minute I just have to suction her. [Laughter] And then they come back and have a chat.*

[P21].

Similarly, this is described in the following excerpt:

*But then I feel more at ease now in the company of people, who are either in the same position as me or people who have already been down that road. I can be far more honest with them than I can with people that I might have been friends with for years. [P18].*

Parents felt that only parents in situations similar to their own could truly understand the daily challenges they faced so these parents became part of their 'new life' and part of the 'new' normal they strove to create.

Parents predominantly spoke about creating and achieving normality in or around the family home. Many mothers in particular spoke about the efforts they made to maintain the home as normal as possible. However the home frequently needed to be altered to accommodate the needs of the child. Moreover the presence of equipment regularly intruded the home serving as a constant reminder that 'normal' life was undoubtedly altered and often compromised that normality:

*Probably one of the things they wanted to get us was a hospital cot. We didn't want a hospital cot. They wanted a hospital cot because it's easier for people's backs. And I wanted to keep things as normal as possible. I didn't want him to come home to a hospital. [P16].*

Nonetheless, parents believed that the home environment benefited the child and family and represented a safer, more normal environment than the hospital. Providing care at home minimised disruption to family life and normal routines such as school. It also allowed easier access for extended family members. Additionally, parents gained a sense of control and could focus on performing caring routines in their home environment.

All parents found inevitable hospital admission more stressful than providing care at home; compromising that normality they had created and sought so ferociously:

*People think you get used to being in the hospital, and you don't, it gets harder because you're away from your other children, it's breaking your heart. You're in an environment where you're sitting on a kitchen chair, you can't sleep, and you're providing his care. [P14].*

The 'new normal' that marked family life was completely disrupted during periods of hospitalisation. Siblings were frequently shuffled among extended family. All parents felt mentally and physically drained during periods of hospitalisation as they strove to care and protect their child in this environment which was not the normality that parents had created for their child in the home. Parents also feared that in this environment their child would come to harm for example, pick up a hospital acquired infection while in hospital. Parents unanimously stated that they continued to provide the majority of the care while their child was in hospital. Furthermore, parents outlined how they were unable to get a break when their child was hospitalised: *"It is awful. I figured out bring food with you because you don't get out for food."*[P16]. The hospital environment was viewed as extremely busy and parents feared that their child would be left alone or would not receive the necessary care if they were not present.

In the constant strive for normality for their child and family, parents appeared to prefer to care for their child at home whenever possible, even though the pressure on parents increased as the child became sicker as outlined by two parents below:

*We keep him out of the hospital at all costs. And we've done it on numerous occasions, even when it was unsafe to do so. Because every time he has ever been in hospital for any length, he has come out sicker than when he went in. [P18].*

This was also outlined in the following statement:

*He got quite badly sick and we put the NG tube down. I knew, if I phoned (doctor's name), she would say, bring him in. And we were like, no; we are not bringing him in. We can manage this. And it was getting scarier. And he wasn't great. We were getting the fluids in and like we knew he was border line. We knew we were walking that line. [P16].*

As the child became more unwell at home, parents greatly appreciated when healthcare professionals supported them in their decision to care for their child at home:

*We got past it, and then I contacted (doctor's name) and told her everything..... She said that's exactly what they'd have done in hospital. And I was then happy that I'd confessed, that I'd confessed after the fact. [P16].*

This was also outlined in the following excerpt: *"other healthcare professionals have quietly acknowledged that they think it has stood to him."*[P18]. In avoiding the hospital and having support

from understanding healthcare professionals, the parents' felt more able to valiantly strive for normality.

As parents strove to maintain normal routines and create a 'new normal' they became adept at organising, including arranging and fitting in appointments, preparing information for healthcare professionals (such as summarised medical histories) and being well prepared for contingencies. Care needed to be managed twenty-four hours a day and for some, this involved significant interruption to sleep. Lack of sleep and constant pressure made managing this new life incredibly difficult: *"we just make it work on a day-to-day basis, but it is really hard."*[P10]. Parents developed expertise in managing the care of their child in addition to responding to the needs of the whole family. Thus, managing this 'unexpected life' also had at its core developing the skills and expertise of parenting in a different way. It was indeed a journey overtime hence 'Becoming the expert in their child's care' arose as a subtheme given its centrality to 'Managing an unexpected life'.

#### **5.4.2 Becoming the 'expert' in their child's care**

This subtheme relates to how parents transitioned from 'learner' to 'expert' in their child's care. Parents clearly indicated that their confidence and competence in their ability to care for their child increased over time. Furthermore, they outlined how carrying out technical care became part of their normal routine:

*Then you do bits around the house, and then at five o'clock you start nebs, chest physio, and the cough assist machine. Then it's back on the feed again. He's got better now. The nights were dreadful before. They are better now. [P4].*

The fear which was associated with learning to care for their child dissipated in time.

Often associated with lack of faith in the 'medical system', they began to develop belief in their own abilities, realising that they could listen to their intuition, learn from their child and trust their own experience of caring for their child:

*Like after his first smile I remember saying, I'm never going to trust any of those doctors again. I remember saying to (child's name), no more listening to the doctors about what you are going to do. I'm going to watch you from now on. So that was very helpful because he'd be smiling. You'd bring him out for a walk and you could tell that he enjoyed the walk. [P10].*

As parents transitioned from 'learner' to 'expert' they developed an increased sense of competence in carrying out often very complex procedures that previously caused them great anxiety. So for

example, the magnitude of the responsibility for procedures such as changing a tracheostomy was previously viewed as a daunting task:

*I was nervous about everything at first, and trying to do it all for the first time, and even getting your confidence up to the stage that I am at now. I'm at the stage now that I literally can change a trache in my sleep. [P23].*

Thus, with increased experience and confidence such procedures became normalised within their new life and different parental role, as they developed expert management and caring skills. This is illustrated in the following statement: *"I woke out of my sleep one time and changed his trache and I couldn't even remember doing it. So I literally can change a trache in my sleep."*[P23]. The emotional burden of carrying out the complex care required by the child also eased, with either a growing sense of acceptance and/or a perception that carrying out the procedures had become easier over time.

However, parents wanted to see themselves primarily as parents and did not want their relationship with their child to be defined by the nursing activities they undertook. They described a sense of resentment at the way in which their nursing role sometimes dominated their parenting experience: *"Sometimes I feel like (child's name) nurse and not his mammy. I do, and I hate it. I wish I didn't feel like that."*[P4]. Parents had to learn where the boundary was for them between parenting and nursing roles: *"but one thing we always refused was how to learn to pass the ng tube, because that went beyond... I needed some sort of a cut-off between mummy and nurse."*[P7]. At times parents struggled to create boundaries. For example, some parents had a choice in taking on responsibility for carrying out the procedures, whilst others did not. Where parents did not have a choice; this was sometimes because of limited service provision locally. However the majority of parents were in two-parent families and, of those; many shared the responsibility for the procedures to some extent with their spouse:

*I did her oral medication, and he would have done the pump medication...I was too worried because making the medication up was very, very hard. It had to be precise. So then (husband's name) took over, and he was brilliant at it. He could do it with his eyes closed. [P20].*

Occasionally fixed roles had been assumed, in other cases, parents took turns with the different roles required. Where responsibility was shared with their partner, parents greatly valued this.

In becoming the 'expert', parents developed an intimate knowledge of their child's care needs which were perceived to be nuanced, unique and different. However, the requirement to be able to adapt

to changing needs was at times overwhelming and could (as parents saw it) undermine their sense of competency and know-how. Another issue for parents was the perception that healthcare professionals on occasion did not recognise them as 'expert' in their child's care. Parents indicated that they thought their expertise should be recognised, but this was not always the case. On occasion, healthcare professionals as parents saw it positioned themselves as experts, without taking into account parents' views which led to parental frustration:

*She doesn't listen. It's frustrating, because it's hard enough. We're not trying to tell anybody how to do their job, but we're saying, would you stop and listen to us first? And understand why we're trying to say what we're saying. [P15].*

Additionally, parents felt that legitimate health concerns raised by them were often dismissed, and parents had to challenge healthcare professionals to be taken seriously. As a component of becoming an 'expert', parents gained confidence in their ability to question healthcare professionals' opinions:

*I said, I think he has a dislocated hip. And he said to me, no, no, no, that's just his... he's very hypotonic, physiotherapy will help. So I just looked at him and I said I'm not leaving until you get him x-rayed. I said, it won't be too hard, radiology is downstairs. And he didn't really take kindly to that but he organised it. And I went down to radiology, and the radiographer said to me, this is just to rule out a dislocation, is it? And I said, well it's to see if he has a dislocation. And again, because I've no experience whatsoever, I just knew as a mum. And he came back and he called me in and he said, yes, this is a very significant left hip dislocation. [P10].*

However, parents frequently stated that they had to be persistent with healthcare professionals about their concerns.

In becoming the expert, parents regularly adopted the role of information provider when it came to their interactions with a number of healthcare professionals. They had to continuously repeat information, some of which related to the rarity of the child's condition, but also at times due to poor communication between healthcare professionals. This was particularly evident in the Emergency Department. Parents often felt resentful when they were expected to adopt this role:

*I understand if they're not qualified on that condition. But I mean even to know a little bit about it would help. They don't have to know everything. Even a little bit, because I have to go back through all the history every time I go into A&E, because it's a new doctor. Every time you go into A&E it's a different doctor, and they're asking what's (name of syndrome)? I've even been asked by some doctors, how do I treat a child with (name of syndrome)? [P22].*

The sense of resentment is tangible in the following excerpt:

*We're in the metabolic clinic every six months. So when they're writing their notes, they put, (child's name), (name of syndrome), and they'll write little notes in from the past, and what's happening now. So even if they read the last notes, they would know a little bit, do you know what I mean? But they don't want to even do that. [P22].*

Parents found this mentally draining as they had to continue to repeat harrowing information regarding their child's condition, and having to do this often contributed to a lack of trust developing in the abilities of a particular professional/or professional grouping.

Although parents inadvertently became the expert in their child's care, they needed education /training to develop the new skills required by the changing needs of the child. As well as education parents indicated the need for ongoing support in managing the ever changing care needs of their child. Whilst parents reported periods of relative stability in their child's condition, there were also periods of illness and decline, when they needed 'expert professional support' to enable them to continue in their parenting role. However, this support was not always readily available. Parents recounted how they fought often on a daily basis to access services and support systems in order to manage to care for their child and ultimately manage their unexpected life. Thus 'Fighting for your child' arose as a subtheme as it was an integral component to 'Managing an unexpected life' as parents saw it.

### **5.4.3 Fighting for your child**

This subtheme relates to the parents' determination to fight on their child's behalf whether this meant securing a second opinion, exploring treatment options or securing optimum levels of care. Both mothers and fathers fought intensely on their child's behalf. Fighting was perceived as essential in order to manage to care for their child. Parents stated that they had no choice other than to fight: *"you have to fight."*[P3]. Parents, however spoke of the mental and physical exhaustion that they suffered as a result of fighting:

*The battles that we have had with getting him diagnosed and getting treatment for him, and getting help, has probably taken their toll far more than the actual condition has. It's just what it has taken out of you mentally. It's so draining. [P18].*

This was also reiterated in the following comment: *"that's what I find really drains you because sometimes you just can't be bothered, but you have to fight."*[P21]. In addition, the need to fight on their child's behalf was perceived as relentless:

*I suppose the hard thing is just the constant fight for everything. It's very, very hard. Sometimes you just want to be a mum, but you've got your pile of paperwork or your phone*

*calls to make or your emails to send, and you always have to be a bit ahead of the game. Like, you can't drop the ball. That's hard. [P21].*

Parents described a sense of resentment and frustration when fighting on behalf of their child overshadowed their parenting experience and marked everyday family life.

Parents frequently felt alone and unsupported in their fight to secure services for their child. This was clearly evident in the following quote: *"Not one person has fought his corner for me. Not one."*[P23]. This feeling was also repeated in the following excerpt:

*You do fight for everything. You fight for his hours. You feel like there aren't many people for you. There definitely aren't many people for you. I could count on one hand whose there for you. [P4].*

Moreover, parents were required to go to enormous lengths to prove that they needed help. This is described in the following scenario:

*He was asleep. I woke him, stuck him on his feed even though he wasn't due it, stuck him on his vent and plonked him in the corner. So every time he moved, the ventilator came off. Now, he's full care but he can be great at the same time. But he needed to see the reality, and the reality is this is what it's like. [P3].*

Additionally, parents reported that healthcare professionals frequently made them feel guilty as they fought to secure services for their child. Parents intermittently spoke about healthcare professionals who advocated on their child's behalf or who engaged in the fight with them. Parents felt a sense of relief when they were supported in their fight to secure services for their child:

*I just burst into tears at the table. And I said you're the first person that has sat here and actually offered me help. You're the first person that has said I'm not making you jump through a, b, c and d before I'm going to help you. [P18].*

This sense of relief is palpable in the excerpt.

Parents had to learn to fight in order to manage to care for their child. Parents appeared to acquire the necessary skills in order to advocate effectively as they proceeded on their care journey:

*I said - and it was probably just to put the point across - I said, it's been of huge value to me and my own headspace and mental health. I think as soon as she heard the word mental health, she went, Oh God, we'd better give her some hours. And it's terrible to have to use things like that. But it is my mental health. [P17].*

Similarly, another mother outlined her approach: *"be nice. I'm not saying be a two-faced (curse) cow. Be nice, but at the same time, don't be a pushover."*[P3]. Parents assumed the role of

advocate even when taking on such a role was not comfortable or was not in keeping with the way they usually acted. Parents expressed that they felt fighting for their child was difficult because they could not afford to alienate influential healthcare professionals as this could impact negatively on their child and the services they receive. This in turn made fighting for their child even more difficult as it was a balancing act for parents.

Additionally, in fighting for services, parents encountered major gaps in service provision. In order to access services, parents felt that they had to fight the system. In all service areas, parents reported that the bureaucracy often required large amounts of paperwork. The repetitive assessments that parents had to undertake were time consuming and frustrating. This impacted significantly on their time in addition to daily care giving duties:

*every year you come up to review and they try to take away his care, try to reduce his care package, and that's where my battle is every year, his condition hasn't changed, nothing has changed, but they will do their best to try and take the care off him. [P16].*

Withdrawal or the constant threat of withdrawal of services was a source of constant anxiety for parents. Unnecessary bureaucracy caused additional and unnecessary stress. Parents also expressed frustration with the amount of time, energy and personal resources that were required to ensure that their child's needs were met. This ultimately made managing this new life extremely difficult.

In fighting parents talked about encountering major gaps in service provision. Parents were particularly concerned with the lack of therapeutic interventions provided for their child. There appeared to be a marked disparity between what the child needed and what the services could provide: *"He hasn't had a physio appointment in over four years."*[P23]. Moreover the services provided were frequently not tailored to meet the individual needs of the child: *"her main need where physio is concerned is chest...the physio actually doesn't do chest physio, and I thought, my God, what use is that to us?"*[P12]. Parents suggested that not only was there insufficient funding to operate services, but there were insufficient healthcare professionals to deliver such services, so for example: *"the speech and language therapist left last year. There's still no speech and language therapist. There's an emergency number if you need it. So that's a joke."*[P12]. Therapists were not replaced when on maternity leave, annual leave etc. and so no service was available to the child during these occasions, making the case for parental 'fighting' even stronger.

Another influence on parents' perception of the need to 'fight' was the apparent geographical inconsistency in the provision of in particular of out-of-home respite services. Moreover, in many cases, access to respite services in known established providers appeared to be on an ad-hoc basis:

*So the doctor sent in the referral, and it must have been about a month or so later - we got a letter back listing what (name of service provider) does ... It cares for children... end-of-life and...and then the next bit (child's name) doesn't currently meet this criteria, so we won't be going any further with this referral. If things change, please do get in touch. And I was shocked when I opened it, because I thought when you put the referral in, I thought they'd come out and visit us, and see our situation, and meet (name of child). So I was a little bit disgusted that they didn't even come out and meet him. [P17].*

In addition and as a result of their fighting, some parents expressed dissatisfaction with the inappropriateness of some settings in which respite care was offered:

*We were offered respite here and we went there on three occasions. But, it's not for us because it is a respite facility for intellectual disabilities, and it's both an adult and child facility. So, half the month is devoted to adults and half the month is devoted to children. But I don't feel you can operate a service that's designed for both adults and children, even in terms of the physicality of the building. There are no bright colours. There are no murals on the wall. There are no toys. Not one toy. And I can't send my son, that's a very, very sociable... Loves Company, loves bright colours, loves music. I can't send him somewhere like that and leave him there because it doesn't do us any good. I'd be as well off without it. [P18].*

Parents who had access to out-of-home respite care indicated a sense of both guilt and relief when they were granted it and started using it. So as in the above case parents' ambivalence to using respite was compounded if they were dissatisfied with the services available to them. Even when the fight yielded suitable out-of-home respite, this was not always problem free for parents. It seemed to in some ways make parents feel that they had let their child down, but in fact they needed it to refresh themselves with the physical strength to continue caring.

Securing in-home respite also posed many challenges, instigating battles for parents. The complex care required by the child made getting a suitable healthcare professional to care for the child very difficult. Often a qualified healthcare professional was required but their availability was limited. This resulted in in-home respite hours being organised around a time suitable for the professional carer to suit the needs of the carer rather than the needs of the parents:

*And as well as that, a lot of them have to be working in the acute services. So they'd be working three days a week, so they wouldn't be available to you those days. So you are*

*curtailed with their availability, but at the same time they are my saving grace. They are the reason that I can run things normally. [P10].*

The voluntary sector played a significant role in the provision of crucial respite services in the child's home. All parents viewed in-home respite care in a positive light in that it enabled them to manage their particular situation whilst although their perception varied, ranging from viewing it as enhancing family functioning and being critical for their survival. Such outcomes gave impetus and credence to the fights that parents became involved in.

Many parents spoke of their 'battle' to secure equipment they believed their child required. Parents waited months for buggies and suitable seating for their child:

*(Child's name) was 18 months old before she got a chair or anything, and we'd sit her in the corner here, and to this day we still sit her in the corner sometimes, because it's just purely out of habit. [P5].*

This made caring for the child very difficult. Many parents declared defeat, sourcing and purchasing their own equipment. Parents also described that lack of information contributed to protracted fighting that was often unnecessary:

*And yet there are all these things out there that you discover. Oh my goodness, I should have applied for that or I should have done this and there's that grant. You do get told in time about certain things but an awful lot you don't get told about. It's only through somebody else saying something. Some of the parents maybe say we got this because we applied for it or whatever.... [P6].*

Information from other parents was considered a particularly important source of information on services and entitlements, serving as vital ammunition in their ongoing battles for their child. However, parents tended to collect information in fragments, which increased the likelihood of information gaps.

So 'Managing an unexpected life' was challenging and an always unfinished business for parents that required adaptation dependent on the child's condition. Given that these children had progressive conditions, this was not surprising. Three elements were therefore central to parental management those being 'Striving for normality', 'Becoming the expert in their child's care' and 'Fighting for your child'. Family life was disrupted and chaotic; however, parents employed important action strategies to enable them to manage as efficiently and effectively as possible for their child and family. The third and final dominant theme 'The ship is going down' will be outlined next.

## 5.5 The ship is going to go down

The third theme that emerged was the parents' often never verbalised but ever present experience of living with the knowledge and uncertainty that whilst death was inevitable, they did not know when it was likely to occur:

*We're all on board the same ship, and we're all going in the same direction. We don't know what direction the ship is going but somewhere the ship's going to go down. Everybody that's on the ship is going down with it. There's no getting away from that. It's scary. [P15].*

Parents had a sense that their child's death was inevitable from the period of diagnosis or recognition of a life-limiting limiting condition. This sense of awareness continued through the time period where parents began to manage. Thus the third theme 'The ship is going to go down' underpinned the other two themes 'Starting out in haziness' and 'Managing an unexpected life'. The theme 'The ship is going to go down' incorporated two specific areas which arose as subthemes those being 'Living one day at a time' and 'Preparing for Armageddon'. See Figure 8.



**Figure 8: Diagrammatic representation of Theme 3**

All parents voiced fear and sadness when they contemplated their child's future. The interviews suggested reluctance, on the part of most parents, to look too far ahead into the future:

*She says she doesn't know how long he has left, but she reckons he definitely won't make it past the teens. I just can't imagine my life without him. So when that gets said to me, I'm like, no, he'll be grand he'll be here. He is still here, he is still happy. That's the main thing at the minute. [P8].*

Parents revealed feelings of uncertainty when they considered life in the future:

*It's just totally you can't fathom it. The prospect of losing your child is just so overwhelming; I can't even contemplate what it will be like to actually go through it. It scares me to death. And you wonder how you will ever live after that, but I know that you have to. [P18].*

Parents could not avoid anticipating what was to come. All parents reported feelings of helplessness, knowing death was inevitable, albeit unspeakable.

Parents felt uncertain about making long-term plans for their child and family: *"because you don't know what the future is going to hold."* [P22]. On the contrary, having a child with a life-limiting condition gave parents the desire to maximise the opportunities available to them:

*So our lives now are about trying to move on and make memories. Go places, take her to places that she hasn't ever been to before, and that she might never get to. For me, my whole life now revolves around making memories. [P20].*

Parents learned to really appreciate the quality of their child's life, abandoning the idea that quality is measured in terms of life expectancy:

*She's very clever in her own way. She's cleverer in a lot of ways than what the doctors said or thought. She knows the Mr. Tumble signs. She knows the Incy Wincy spider signs. She knows the wheels on the bus. She can do all those. [P11].*

Parents also focused on the meaning of their child's life. Moreover, they spoke about shaping their child's life in a positive way:

*So, we've seen her place in the world now, she went to preschool and she goes to school, and she has friends now. Okay, it might be different, but I suppose she's brought lots of new friends into our lives that all have different struggles as well. [P21].*

Parents made every effort to ensure that every moment was special for their child.

Yet parents voiced concerns regarding the strain of living with uncertainty in many areas of their life; however of primary concern was the uncertainty surrounding their child's trajectory:

*And the hardest thing for us as well was we were told her condition is life-limiting, and they can't obviously put an age on it, so you don't know how long she's going to be here or what's going to take her, or anything like that. [P21].*

Uncertainty surrounding their child's trajectory almost became a routine part of life from the moment of diagnosis or recognition of a life-limiting condition. Parents described the journey as lacking any sort of roadmap or guidance. Moreover, they frequently described how their child had outlived original prognostic predictions:

*We were told he wouldn't make it past 6 months, he wouldn't make it past 2, he wouldn't see (curse) this and he wouldn't see that... Every couple of years this went on where they just pushed out the date. [P2].*

As a result, parents appeared to have a degree of scepticism regarding medical prognoses.

In addition, parents perceived that services were provided or in some instances not provided for their child based on medical prognoses:

*And we took him back home. We weren't expecting to have him that long so we weren't. And I suppose a few issues when we did get out. We got home, and he had no medical card, which people were very surprised with, because we had taken home a very ill baby. [P19].*

Moreover, it often took a considerable length of time to secure vital services for their child:

*I suppose, physio, and a lot of these things weren't put in place, because he nearly had to prove his worth. It was like oh well, I'm here now ten months, and I've earned these things, where a lot of this stuff at the start was very much, well; enjoy him while you have him. [P19].*

This made caring for their child particularly challenging for parents.

Additionally, parents described how there were times when their child defied expectations and survived episodes of life-threatening deterioration: *"he did make it and he's here now, you know, when he shouldn't be, nature says he shouldn't be."* [P15]. Parents regularly indicated that survival from episodes of life-threatening deterioration set them apart from other children and their families: *"very few actually survive so she is really defying the odds at the moment, and they're also learning from her because there is no one else."* [P8]. Repeated survival from such episodes made the threat to the child's life uncertain, illustrated by ideas about the child being 'strong' rather than fragile: *"he was showing us that he was a little fighter, a little soldier after everything he'd been through."* [P10]. Parents appeared to deal with the uncertainty in their lives in a pragmatic way, living one day at a time. Thus 'Living one day at a time' arose as a subtheme as it was a key component for parents who were aware that the 'The ship is going to go down.'

### **5.5.1 Living one day at a time**

The strategy of living one day at a time appeared to allow parents to forget and not dwell on the inevitable that 'The ship is going to go down'. This sentiment is echoed in the following quote: *"our motto from day one has always been we're going to live in the moment."* [P18]. Living one day at a time was therefore experienced as a coping mechanism and as a necessity. This was not to say that

parents did not consider their child's future. While they focused on living, their narratives revealed that thoughts of death were never far away:

*I do worry about the future. I worry about the time when he will not be here. You don't know when it's going to happen. Nobody knows. I would just like that certainty, in my life, but it's just, it's not there. [P7].*

Indeed, the fear and realisation that their child's death was inevitable was very real: *"I know the time's going to come when he won't be here with us."* [P18]. However, adopting a one day at a time approach helped parents avoid concentrating on these possibilities. Additionally, this approach also appeared to empower parents to stay positive.

Parents without doubt described positive feelings of parenting a child with a life-limiting condition: *"He changed my life, yes, 100% for the better."*[P12]. Parents admitted to seeing life in a different light and changed their priorities:

*I can honestly say maybe life wasn't better before, in some respects, because I don't think we appreciated things. We didn't. And I think we've learned lessons now in life that we never, ever would have learned if we hadn't had (child's name). [P18].*

Parents described how siblings became more sensitive and compassionate:

*He just accepts him for whom he is and it's just the most empathetic relationship. It's gorgeous to see that I reared a child so sensitive. And what happened in our whole family situation has made him the kid that he is because he's just so natural with him. He'd say I don't care if he doesn't talk, Mum. [P10].*

The desire to remain positive was paramount, but often very challenging as the journey was revealed to be complex and unpredictable.

Family life was characterised by unpredictability and uncertainty, with parents struggling to maintain as normal a life as possible for all family members:

*Even if I said we'll go out tonight at eight o'clock, well if he was sick or had a seizure at that time you can't go, so you can never say I am doing something. We have a different life. [P2].*

Family activities and outings, including holidays were restricted because of the unpredictable nature of the child's condition. This was frequently a source of guilt for parents:

*It restricts where you go and it restricts where the boys go, but that's a way of life for them now, they've accepted that. They have just learnt to accept it, which I suppose isn't fair either, but there's nothing you can do. [P7].*

All family plans were subject to change depending on the child's condition:

*We have had to cancel so many things. He is that unpredictable. If we are planning a day out, we don't tell the boys because they get disheartened if it's cancelled; we just go. If we're good to go we go. [P7].*

However, preparing for unpredictability appeared to be a mechanism whereby parents tried to avoid being taken by surprise and instead gained some control of their feelings of uncertainty.

Although parents expressed the desire to live one day at a time, it was also essential to contemplate decisions regarding their child's future:

*We've had some very difficult decisions to make. It took us about three or four weeks to come to the decision that a DNR is what we want for her. Yes because it's really not an easy decision to come to. [P5].*

Out of sheer love, many parents decided that if their child's condition deteriorated, they wanted to prioritise their child's comfort, acknowledging that the duration of their life was limited:

*And at the same time, if we decide not to have a DNR, we're doing that for our own reasons and not for her well-being. And at the end of the day she will make her own mind up, as she's proved several times already. [P5].*

Additionally, healthcare professionals often endeavoured to prepare parents for what they perceived as an inevitable outcome:

*And so a couple of months after he had that episode, we were like, oh, we've an appointment with (doctor's name), we have to go and tell him how great things are, we said to him, he's doing really well, he hasn't had a chest infection, and he just turned round and said to us, I don't want to come across as a wet blanket but there's tough times ahead. [P5].*

Yet, parents often believed that the child's survival was determined by the child's own will and fortitude.

However the parents desired choice and had definite ideas about what interventions they believed were appropriate for their child:

*I said if anything happened to him to make him more complex than he already is, or his life more limited, that I would consider maybe not putting him through anything if he was going to come out worse. [P17].*

Some of these decisions shifted and changed, but having the choice was very important to the parents:

*So that was the first question I asked the doctor. I said is his brain damage going to be worse now, after what happened? And he said, we can't tell you. We can't predict that. But then, when you're in that situation, you just have to do everything you can do save him. [P17].*

Decisions were made easier when parents trusted healthcare professionals caring for their child and perceived them as caring. Parents needed time and appreciated when they were afforded the time when faced with challenging decisions: *"they didn't push me. They said, there's no hurry on it, just have a think."* [P22]. Parents, however were not always afforded this time: *"we weren't given enough information. We weren't given enough time to decide. We weren't given support."* [P9]. Parents became more protective, defensive and adamant that all aggressive treatments continue if they felt pressurised into making a decision:

*I was on my own with her, after being in A&E since the night before, she arrived with her team and some students, and in front of them all asked me about my decision about her end-of-life care. I complained .... I said, look, I understand she wants to have something in place, but the way she asked me was not nice. I said do you know what, just for now; just write down that I want everything done. [P21].*

Parents believed that discussions regarding end-of-life decisions should be raised in a sensitive and timely manner.

Notwithstanding the care demands required by the child the periods of stability were perceived as a time to enjoy. Periods of stability were comforting and allowed some distance from thoughts regarding death. During this time parents focused on living one day at a time and spoke of putting their energies into making the best of the time they had together as a family:

*Yes, and the way we see it, at this moment in time, he's as well as can be expected and we've got to go along with that. And that's the way we live, you've just got to go for it. [P14].*

Parents often focused on their child's immediate and current well-being. Nonetheless, in the midst of tentative security, parents often feared that there would be a crisis ahead:

*It's very hard not to think about it. You do try to put it at the back of your mind. In the evening time, when she's gone to bed and I sit down, that's when my brain goes into overdrive because when I'm with her during the day I'm too busy to be thinking, but, yes, it is difficult because you don't know how long you have with her and something could change so fast. [P8].*

The situation was compounded as parents had no idea when a crisis could occur or how long the relative stability would last. Parents were always anxious about the end times and remained constantly prepared for something to happen, although this varied in intensity and depended on

their child's condition: *"you have to prepare for Armageddon. Armageddon might never happen but you have to prepare for it."*

### 5.5.2 Preparing for Armageddon

This subtheme relates to the need to be hypervigilant regarding the child's condition knowing that 'The ship is going to go down'. Against a backdrop of constant fatigue and emotion parents described the need to remain constantly alert for changes in their child's condition: *"you need to be tuned in; you need to be ready in the back of your mind going, what if, what if, what if and that's the way you live."* [P15]. These changes could happen suddenly and with little or no warning:

*So he can change that fast. So you're all the time worried. He's just fragile, he's a fragile kid. You could sit him there and he could be watching TV and you could come in two minutes later and he'd be gasping for breath, so in that sense you know he's tough work.* [P2].

At times there were long periods of stability in the child's condition and other times, crises came quickly or repeatedly. Regardless, all parents spoke about the uncertainty that characterised their day-to-day lives.

Parents often feared that healthcare professionals caring for their child would fail to recognise subtle changes in their child's condition which could lead to acute deterioration:

*You just have to know him so well. Because he only gives you teeny little signs...tiny little signs... If you don't pick up on those signs its tough (curse) because he's just going to do something. He is going to deteriorate.* [P3].

Parental concerns regarding the competency and experience of professional carers, particularly in the home were described:

*We're happy enough to leave her for a couple of hours, but no. She's a care worker. She looks after mostly elderly people. So she's not used to children too much, but she does her best. You'd be happy when you get home. Now she looks after him fine. I'm comparing her to (nurse's name). She's an expert, like. There's no comparison. He goes downhill extremely quickly. So we need a person that would recognise that there is a problem and act on it.* [P1].

Parents were often reluctant to leave their child in the care of others in case there was a sudden deterioration. Full knowledge of the child's condition and associated care needs were considered essential to be able to respond immediately in times of emergency, which were mostly unpredictable and rapidly changeable. Parents above all valued healthcare professionals that were consistent in their child's life: *"we've had the same nurse since he was three months old, and she's just brilliant."* [P17]. Parents described feeling comfortable and more at ease when they had

developed trust or faith in the healthcare professionals caring for their child. Furthermore, parents felt comforted when they could exercise control in maximising their child's quality of life, for example by minimising exposure to respiratory tract infections.

Nonetheless, parents were constantly reminded of the fragility of their child's life: *"my two friends, in the last six months, seven months, have lost their two kids."*[P18]. The deaths of others frequently provided a context for thoughts about death:

*Yes, it is very hard. There have been quite a few children that have passed away in the past seven months. It is very upsetting when you hear it, and especially when you know them. One little boy got sick one day and five days later he passed away. He just couldn't pull through.* [P8].

Parents often found it emotionally challenging to hear of the deaths of others:

*You're sad for them and you're sad for the family. Then you think, God, the sadness of this world, of these children, the poor little pets, all they go through the struggle. Their parents are struggling from day one, having them. They've done their best and then they're just taken away from them. It's hard to see.* [P4].

Parents spoke about psychological coping strategies such as avoiding negative thoughts and feelings:

*I've kind of pushed away from them for a while now again. I want to go and talk to every mother who's lost a child and let them talk and have a cry, but I can't do that.* [P4].

Repressing thoughts of death and dying was viewed as a means to handle distress and sorrow.

Parents often found it difficult to talk about their feelings, concerning their child's death. Yet they had definite thoughts regarding their child's end-of-life care:

*If I can avoid it at all I don't want him to die in hospital and I don't want him to die in a hospice. I do want him to die at home. But I want to be supported. I don't want to be sitting here, going, oh my God, is he in pain now?* [P18].

Some parents had tried to talk about end-of-life and death with family members, but felt their concerns were pushed away:

*Like, there's a lot of things, I can't talk to even my parents about her because, with my parents, all you get is, shush, she's doing fine, she's not in hospital. That's their attitude. Yes, and like that, I suppose its denial too; they don't want to accept there's anything wrong with her.* [P8].

Moreover, many parents did not want to burden their family with their concerns. The parents often prepared in their minds by imagining different scenarios:

*I'm more worried about the future because look, he will die at some stage, he's not going to bury me like – we know that – but my point is she'll be devastated and what will it do to us as a family? [P2].*

Healthcare professionals were frequently a vital source of support. A number of parents recalled conversations where healthcare professionals had communicated in a sensitive and compassionate manner. However, parents often lacked confidence to ask questions and appreciated when healthcare professionals adopted a proactive approach with regard to assessing information needs and providing information. Parents expressed the need for reassurance that they would be supported during end-of-life care:

*I couldn't go through that. Can anybody guarantee me that that won't happen with my son? And he's only six, and I know for a fact, if you come back to me in two years' time, he's probably not going to be here. [P18].*

Additionally, future withdrawal of support was a source of anxiety for the parents:

*All the people that's involved, the nurses, and all the people you've made friends with and you've made part of your life. That's all taken away from you too. Everything's taken away. It's not fair, really. It's just cruel. [P4].*

The need for support at every stage of care, from diagnosis to post death was reiterated during the course of the interviews.

Thus living with the knowledge of certain death at an unknown time was extremely challenging for parents. Parents had a sense that their child's death was inevitable from the period of diagnosis or recognition of a life-limiting condition. This sense of awareness continued through the time period where parents began to manage. Thus, this theme 'The ship is going to go down' underpinned the other two themes 'Starting out in haziness' and 'Managing an unexpected life'. The knowledge of certain death at an unknown time, often remained in the background, but occasionally came to the foreground for various reasons such as deterioration in their child's condition or a conversation which triggered thoughts of death. Two elements were central to parental management knowing 'The ship is going to go down those being 'Living one day at a time' and 'Preparing for Armageddon'.

## **5.6 Overview of quantitative findings**

As previously stated in order to examine decisional conflict and family-centred care, quantitative data were collected from the parents using two standardised measures. The standardised measures used for this purpose, were the DCS (O' Connor, 1993) and the MPOC-20 (King et al., 1995). Data relating to respite care and sociodemographic variables were collected from the parents using a

demographic questionnaire developed by the researcher. The following section reports the results from the data analysis undertaken. The sociodemographic data, because they were parent sample characteristics were presented in the methodology chapter. Findings in relation to access to respite care are reported first, followed by the descriptive statistics related to the DCS and the MPOC-20. Relationships between the DCS and the MPOC-20 are then reported followed by an analysis of relationships of each of these scales and selected sociodemographic and respite variables.

## 5.7 Access to respite care

Parents were asked whether they had access to in-home-respite care. All 23 parents interviewed reported that they had access to in-home respite care. Of the 23 parents, n=3 were not utilising it at the time of the interview as they felt that they did not need it. Parents were asked whether they had access to suitable out-of-home respite care. Suitability was determined by parents themselves. Of the 23 parents interviewed, n=15 (65.2%) reported that they had access to suitable out-of-home respite care, whereas, n=8 (34.8%) reported that they did not. Of the 15 parents who had access to suitable out-of-home respite care, n=3 were not utilising it for the following reasons: not being ready, not wanting to use it yet and feeling it was not needed at the present time. Additionally, of the 15 parents who had access to suitable out-of-home respite care, n=4 had access to two service providers. Chi square analysis was used to examine the association between access to out-of-home respite care and living in an urban versus rural location. Assumptions relating to the number of expected frequencies per cell were violated in this analysis, so further results are not presented. Table 8 below cross-tabulates access to suitable out-of-home respite and dwelling location. Notwithstanding the inability to use Chi square analysis to confirm a relationship, observation of frequencies suggests that urban dwellers had better access to suitable out-of-home respite care.

**Table 8: Cross-tabulation of access to suitable out-of-home respite and dwelling location**

Variable		Dwelling	
		Urban	Rural
Suitable out-of-home respite	No	2	6
	Yes	13	2

## 5.8 Decisional Conflict Scale

Decisional conflict was measured using the DCS (O' Connor, 1993). The DCS measures personal perceptions of: a) uncertainty in choosing options; b) modifiable factors contributing to uncertainty such as feeling uninformed, unclear about personal values and unsupported in decision-making; and c) effective decision-making such as feeling the choice is informed, values-based, likely to be implemented and expressing satisfaction with the choice. A detailed outline of the DCS scale including the scale scores is presented in the methodology chapter. Descriptive statistics are outlined below.

DCS total scores ranged from a minimum of 0 to a maximum of 82.81, with a mean score of 28.46 (SD=21.10). Scores on the DCS Uncertainty subscale ranged from a minimum of 0 to a maximum of 91.67, with a mean score of 42.03 (SD=29.35). Scores on the DCS Informed subscale ranged from a minimum of 0 to a maximum of 100, with a mean score of 30.07 (SD=26.20). Scores on the DCS Values Clarity subscale ranged from a minimum of 0 to a maximum of 100, with a mean score of 22.83 (SD=29.33). Scores on the DCS Support subscale ranged from a minimum of 0 to a maximum of 75, with a mean score of 32.79 (SD=27.87). Scores on the DCS Effective Decision subscale ranged from a minimum of 0 to a maximum of 62.5, with a mean score of 18.75 (SD=16.96). The DCS total scale and subscales were internally consistent with all Cronbach alpha coefficients > than .7. Please see Table 9 below.

**Table 9: Respondents' Scores DCS**

DCS Subscale	N	Mean	SD	Median	Min	Max	Cronbach's alpha
DCS Total (0-100)	23	28.46	21.10	26.56	0	82.81	.941
DCS Uncertainty	23	42.03	29.35	41.67	0	91.67	.741
DCS Informed	23	30.07	26.20	25.00	0	100	.868
DCS Values Clarity	23	22.83	29.33	16.67	0	100	.964
DCS Support	23	32.79	27.87	25.00	0	75.00	.834
DCS Effective Decision	23	18.75	16.96	18.75	0	62.50	.823

Mean scores on the DCS scale and subscales provide additional valuable summary statistics around decisional conflict associated with the immensely challenging decisions that parents encountered. Parents reported a generally low level of decisional conflict in the study. In general parents were informed, were clear about their values and felt supported in the decision-making process. The mean score on the DCS total scale was 28.46 indicating low decisional conflict. However, the scores ranged from a minimum of 0 (no decisional conflict) to a maximum of 82.81 which is associated with decision delay or feeling unsure about implementation. The DCS Effective Decision subscale received the lowest mean score, however the scores also ranged considerably. In contrast the DCS Uncertainty subscale received the highest mean score. Again, the scores ranged considerably from a minimum of 0 (feels extremely certain about best choice) to a maximum of 91.67 (feels extremely uncertain about best choice). The DCS Values Clarity subscale was 22.83 indicating that parents were clear on their values. However, the scores ranged from a minimum of 0 (feels extremely clear about personal values) to a maximum of 100 indicating that some parents were extremely unclear about personal values.

## **5.9 The Measure of Processes of Care Scale-20**

The MPOC-20 measures family-centred care within five domains: Enabling and Partnership; Providing General Information; Providing Specific Information; Coordinated and Comprehensive Care; and Respectful and Supported Care. A detailed outline of the MPOC-20 scale including the scale scores is presented in the methodology chapter. Descriptive statistics are outlined below. Scores on the MPOC-20 Enabling and Partnership subscale ranged from a minimum of 1.00 to a maximum of 7.00, with a mean score of 5.49 (SD=1.77). Scores on the MPOC-20 Providing General Information subscale ranged from a minimum of 1.00 to a maximum of 6.60, with a mean score of 3.69 (SD=1.89). Scores on the MPOC-20 Providing Specific Information subscale ranged from a minimum of 2.00 to a maximum of 7.00, with a mean score of 4.98 (SD=1.54). Scores on the MPOC-20 Coordination & Comprehensive Care subscale ranged from a minimum of 1.25 to a maximum of 7.00, with a mean score of 5.36 (SD=1.78). Scores on the MPOC-20 Respectful & Supportive Care subscale ranged from a minimum of 2.20 to a maximum of 7.00, with a mean score of 5.83 (SD=1.47). All MPOC-20 subscales were internally consistent with all Cronbach alpha coefficients > than .7. Please see Table 10.

**Table 10: Respondents' Scores on MPOC-20 subscales**

MPOC-20	N	Mean	SD	Median	Min	Max	Cronbach's alpha
Enabling and Partnership	23	5.49	1.77	6.00	1.00	7.00	.949
Providing General Information	22	3.69	1.89	3.50	1.00	6.60	.870
Providing Specific Information	22	4.98	1.54	5.00	2.00	7.00	.856
Coordination & Comprehensive Care	23	5.36	1.78	6.00	1.25	7.00	.964
Respectful & Supportive Care	23	5.83	1.47	6.40	2.20	7.00	.945

Mean scores on the MPOC-20 subscales provide additional valuable summary statistics about the relative strengths and weaknesses of current CPC service provision for children with NMLLCs and their families. Parents reported a generally positive perception of family-centred care provided. Respectful and Supportive Care received the highest mean score on the MPOC-20 subscales with parents reporting that this is achieved 'to a great extent'. Thus, it would appear that the strongest aspect of service provision is related to the type of interaction and relationship parents have with healthcare professionals rather than any practical aspect of service provision. In contrast, Providing General Information received the lowest mean score on the MPOC-20 subscales indicating that it is perceived by parents to be the weakest aspect of service provision. Interestingly Providing Specific Information received the second lowest mean score. These results highlight the areas of CPC service provision that may require more specific attention (provision of general and specific information). Although Providing General Information and Providing Specific Information received the lowest mean scores on the subscales there is room for service improvement in all areas. The fact that parents according to the MPOC-20 were least satisfied with information provision is significant in relation to decision-making. Information about options, benefits, risks and side effects can make parents feel more informed in decision-making.

### **5.10 Relationships between variables**

The inferential statistics were used to explore four specific areas: firstly the relationship between the results of the two measures and then the relationship between the results of the two measures and three of the variables. The three variables included: education, dwelling location and access to suitable out-of-home respite care. Correlations (Pearson or Spearman) or independent samples *t*-tests (or equivalent nonparametric Mann-Whitney U tests) were used to examine relationships between variables. Pearson correlations were used where both variables were normally distributed,

while Spearman correlations were used where one or both of the variables were non-normally distributed. Normality of distribution was assessed using the Kolmogorov–Smirnov test.

Firstly, analysis was undertaken to see if there was a relationship between the DCS and the MPOC-20 scores. The provision of support can positively influence both decisional conflict and family-centred care. However, there are many other factors that can influence both decisional conflict and family-centred care, thus analysis was undertaken to see if low decisional conflict scores are related to high family-centred care scores. The research design and sample preclude making inferences of causality. Correlations were non-significant between the DCS total score and both of the MPOC-20 subscales: the MPOC-Providing General Information (n=22),  $r_p = -.393$ ,  $p = .07$ ), and the MPOC-Providing Specific Information (n=22,  $r_p = -.416$ ,  $p = .054$ ). Spearman correlations were non-significant between the DCS and each of the remaining MPOC subscales: MPOC-Enabling and Partnership (n=23,  $r_s = -.367$ ,  $p = .085$ ), MPOC-Coordination and Comprehensive Care (n=23,  $r_s = -.306$ ,  $p = .155$ ) and MPOC-Respectful and Supportive Care (n=23,  $r_s = -.368$ ,  $p = 0.84$ ).

Analysis was undertaken to see if there was a relationship between educational level and DCS and MPOC-20 scores. The rationale for this analysis was based on the belief that educational level could influence access to information and thus impact on decisional conflict and family-centred care (Kaplan, Fang and Kirby, 2017). Education level was not significantly correlated with DCS total scores ( $r_s = -.141$ ,  $p = .521$ ). While education level had small correlations ( $< \pm .3$ ) with a number of MPOC subscales, none of the correlations were significant (MPOC-Enabling and Partnership  $r_s = -.339$ ,  $p = .114$ ; MPOC-Providing General Information  $r_s = -.075$ ,  $p = .739$ ; MPOC-Providing Specific Information  $r_s = -.325$ ,  $p = .140$ ; MPOC-Coordination and Comprehensive Care  $r_s = -.297$ ,  $p = .169$ ; MPOC-Respectful and Supportive Care  $r_s = -.409$ ,  $p = .052$ ). Analysis was also undertaken to see if there was a relationship between urban and rural location and DCS and MPOC-20 scores. The rationale for this analysis was based on the belief that geographical location could influence access to services and thus impact on decisional conflict and family-centred care (Bunnell et al., 2017). There was no difference between urban and rural location on DCS scores ( $t(21) = -.103$ ,  $p = .919$ ). There was also no difference between urban and rural location on MPOC subscale scores: MPOC-Enabling and Partnership ( $U = 46.5$ ,  $p = .377$ ), MPOC-Providing General Information ( $t(20) = -.154$ ,  $p = .879$ ), MPOC-Providing Specific Information ( $t(20) = 1.544$ ,  $p = .138$ ), MPOC-Coordination and Comprehensive Care ( $U = 40.50$ ,  $p = .203$ ) and MPOC-Respectful and Supportive Care ( $U = 43.0$ ,  $p = .262$ ).

Finally, relationships between self-reported access to suitable out-of-home respite and both the DCS scale and MPOC-20 subscales were also analysed with independent sample t-tests or the nonparametric equivalent, the Mann-Whitney U test. The rationale for this analysis was based on the belief that access to suitable out-of-home respite could impact on decisional conflict and family-centred care (Keilty, Nicholas and Selkirk, 2018). There were no significant differences between participants who did or did not have access on any of these constructs: DCS total score ( $t(21) = 1.221, p = .235$ ), MPOC-Enabling and Partnership ( $U = 57, p = .845$ ), MPOC-Providing General Information ( $t(20) = -1.477, p = .155$ ), MPOC-Providing Specific Information ( $t(20) = -1.271, p = .218$ ), MPOC-Coordination and Comprehensive Care ( $U = 51, p = .557$ ) and MPOC-Respectful and Supportive Care ( $U = 41.5, p = .222$ ). Although there was no statistically significant relationship between the results of the two measures and between the results of the two measures and the variables the small sample size could have hindered statistical comparison.

### **5.11 Interim comments**

Throughout their care journey parents faced numerous challenging decisions. Decisional conflict was measured using the DCS. Parents reported a generally low level of decisional conflict in the study. In general parents were informed, were clear about their values and felt supported in the decision-making process. However, the scores ranged from a minimum of 0 (no decisional conflict) to a maximum of 82.81 which is associated with decision delay or feeling unsure about implementation. Parents' perceptions with regard to the extent to which the health care services they and their child received were family-centred were measured using the MPOC-20. Parents reported a generally positive perception of family-centred care provided. Respectful and Supportive Care received the highest mean score on the MPOC-20 subscales. In contrast, Providing General Information received the lowest mean score on the MPOC-20 subscales indicating that it is perceived by parents to be the weakest aspect of service provision. Interestingly Providing Specific Information received the second lowest mean score. These results highlight the areas of CPC service provision that may require more specific attention (provision of general and specific information). There was no statistically significant relationship between the results of the two measures and between the results of the two measures and the variables; however, the small sample size could have hindered statistical comparison.

## **5.12 Concluding comments**

This chapter has illustrated the unexpected and uncertain journeys undertaken by parents who find themselves caring for their child who has a NMLLC. Whilst the experiences of parents were unique, in many ways the stories recounted by parents made clear that their journey into the unknown began at the time of diagnosis or recognition of a life-limiting condition. This time period signified a transformation to both their lives and that of their families. As time progressed parents attempted to respond to this transformation, however, this was not without significant challenges. The collective efforts of individual family members, friends, healthcare professionals and service providers helped shape their overall experience. While their interactions with individual healthcare professionals varied, attention was frequently focused on the need to fight with determination for their child's right to have access to services often with little success. Their sense of frustration was apparent, particularly around accessing therapeutic interventions, respite services and services to facilitate home care. The perceived necessity to fight for services exacerbated feelings of loneliness and isolation. Most noteworthy it also added to the experience of living with uncertainty. In addition to the uncertainty associated with the child's condition parents articulated concerns regarding the future availability of support and services to assist them to meet the child's ongoing care requirements. Geographical inconsistency in the provision of services was apparent most notably in the provision of out-of-home respite services. Parents also encountered an array of challenging decisions throughout their care journey. In the next chapter the key findings to emerge will be critically discussed with reference to the initial study aims and objectives and set in the context of the current evidence base.

## **Chapter 6: Discussion of findings: Parents**

### **6.0 Introduction**

In this chapter the key findings to emerge will be critically discussed with reference to the initial study aims and objectives and set in the context of the current evidence base. Thus, this chapter will present a detailed discussion of the qualitative and quantitative research findings from the parents. This will include an integrative discussion. The chapter will begin with a discussion surrounding the experience of caring for a child with a NMLLC from the perspective of parents including their experience of service provision. The discussion following this will concern parents' views of accessing services when caring for their child. Subsequent to this the discussion will provide an insight into decision-making regarding the care and management of children with NMLLCs.

### **6.1 Parents/families experiences including their experience of service provision**

The findings from all three themes provide an in-depth understanding of the experiences of parents caring for a child with a NMLLC. As previously stated the theme 'Starting out in haziness' represented the time period from diagnosis or recognition of a life-limiting condition and marked the beginning of an unknown journey filled with emotional turmoil. This time period signified a key moment in each parent's experience and marked the end of a previously known and familiar world. Likewise, parents of children with life-limiting conditions in other studies revealed that the initial diagnosis is a very traumatic and inwardly personal experience (Collins et al., 2016; Somanadhan and Larkin, 2016). The findings of a study undertaken with parents of infants diagnosed with Hypoplastic left heart syndrome suggest that they are often overwhelmed and confused about how to cope with the apparently ominous nature of their child's condition (Rempel et al., 2012). Parents of infants admitted to neonatal intensive care also described similar sentiments (Hall, 2005; Hall, 2006; Steyn, Poggenpoel and Myburgh, 2017). They reported that the experience resembled being in another world, alien from what they previously knew and experienced.

Parents reported a range of intense emotions such as shock, numbness, fear, low mood and guilt at this time. Sometimes these emotions were coupled with a sense of relief that their child had ultimately survived. Previous research with parents of children with life-limiting and life-threatening conditions identified common parental reactions (Sabzevari et al., 2016; Together for Short Lives,

2013). The findings of a study undertaken with mothers of infants diagnosed with moderate to severe congenital heart disease reveal that they experienced intense fluctuating emotions at the time of diagnosis (Harvey et al., 2013). Upon hearing the diagnosis there appeared to be elements of mothers trying to 'hold on'. They mentioned needing to literally hold or have close proximity to their baby. The experience of having to hand over their infant during surgery and other procedures interfered with this closeness. They described aspects of letting go, a self-awareness process in which mothers recognised that they could not provide all of their infant's needs. Many of the parents in the current study experienced similar sentiments. They likewise realised that they could not provide for all of their child's needs. They also realised that they could not safeguard their child's survival. The emotional impact on parents and the combination of positive and negative emotions were immensely noteworthy at this time.

A complex interplay of factors relating to the internal world of the parents and to the external world, such as the support, compassion and assurance they received determined the nature and the extent of this emotional impact. The parents in this study appeared to have different support systems or means that they used to cope with such a crisis. These encompassed both maladaptive and adaptive coping strategies. Parents appeared to use maladaptive coping strategies to begin with however as time progressed, they predominantly used adaptive coping strategies. These included problem-solving coping strategies and cognitive restructuring. Gellman and Turner (2013) suggest that coping refers to the conscious efforts that are used to reduce the physical, psychological or social harm of a stressful situation and facilitate the interaction of parents with their ill child. The findings from previous research undertaken with parents of children admitted to a pediatric intensive care unit reveal that parents relied on faith, solidarity, spirituality and social network among others (Hayakawa et al., 2010; Robinson et al., 2006). The parents in the present study highlighted the significant impact of supportive interaction with family members and healthcare professionals in helping them deal with the situation. Frankel and Wamboldt (1998) propose that how a child's illness interferes with and impacts a family is determined by the psychological health of the primary parent caregiver. The psychological health of the parent is in turn affected by family functioning. Furthermore the findings of a study undertaken with parents of children admitted to a pediatric intensive care unit emphasised the crucial impact of supportive interaction with healthcare professionals in helping them deal with the stressors of the situation (Majdalani, Doumit and Rahi, 2014).

The manner in which the diagnosis was delivered left a lasting impact with parents able to vividly recall how, when and by whom the information was provided. Poor delivery of information was commonly reported and had a long-lasting impact on parents' ability to adapt and adjust to the situation. This involved overwhelming parents with too much information, insensitive communication and lack of awareness of the significance and impact of the information communicated. Hsiao, Evan and Zeltzer (2007) identified aspects of physician communication that parents perceived to be facilitative or obstructive in paediatric palliative care. The information process itself was a critical element of communication and when the information was experienced as unsatisfactory parents reported feeling frustrated, dismayed and even devastated. Davies, Davis and Sibert (2003) demonstrated that poor communication by paediatricians led to the dismissal of parental concerns resulting in delayed diagnosis of a life-limiting condition with serious repercussions for the families. Comparable findings have been reported in other studies and recommendations made to enhance communication between healthcare professionals and parents (Hill and Coyne, 2012; Lalor, Begley and Galavan, 2008; Lemacks et al., 2013). This study clearly illustrates that poor communication from healthcare professionals at the time of diagnosis is still a concern for parents of children with NMLLCs. In addition, the findings support the literature that poor communication during the time period leading up to and including diagnostic disclosure can result in serious consequences and can increase distress for parents even over the long-term.

Prior to their child's diagnosis, the majority of parents had no previous experience of care provision for a sick child. However, over time and through necessity, parents acquired and developed the skills that enabled them to provide the care that their child required. Many parents provided intensive, highly skilled and complex care shortly after diagnosis. Consistently parents referred to the stress associated with the experience of learning to care for their child. For some, this stress was eased by the support they received from healthcare professionals. This finding was similarly reported in a study undertaken by Brenner et al. (2015) with parents of children with complex healthcare needs. Regardless, parents described the enormity of the caregiver role at this time. They described being both physically exhausted and mentally weary as a consequence of caring. Chronic fatigue and parental exhaustion are reported in the literature associated with life-limiting conditions but not specifically in the time period following diagnosis (Collins et al., 2016; Knapp et al., 2010; Rodriguez, 2009). Parents descriptions of sleep disturbance and in many instances, chronic sleep deprivation in the time period from diagnosis or recognition of a life-limiting condition emphasises the need to address this as a serious problem. The shift in focus towards home and community-based care could explain this finding. In the past, many children would have spent

prolonged periods of time in hospital due to the complexity of their care needs (Noyes, 2000). However, the development of a range of portable devices to support children at home and greater knowledge about the long-term management of childhood life-limiting conditions have meant that children are now cared for primarily at home by their parents (Guglieri and Bushby, 2015; Heaton et al., 2005). Consequently, parents are increasingly taking responsibility for the management of their child's condition early in the course of the trajectory and providing many elements of care previously carried out by healthcare professionals (Glendinning and Kirk, 2000).

The complexity and nature of care provided by parents in this study is akin to that reported in previous research incorporating children with life-limiting conditions (Nicholl and Begley, 2012; Smith, Cheater and Bekker, 2015). The current study, however, also identified the lack of professional support for parents in relation to their role as caregiver which led to parents feeling overwhelmed in the period following diagnosis. There was little evidence of care negotiation between the parents and service providers at this time. Likewise, parents of children with chronic conditions described needing more reassurance and support from healthcare professionals through this time period in order to undertake their new care giver responsibilities and alleviate anxiety (Geense et al., 2017; Rankin et al., 2016). Previous research has highlighted the lack of support for parents of children with life-limiting conditions and complex care needs during the course of the care trajectory but again not specifically in the time period following diagnosis (Coad et al., 2015; Woodgate et al., 2015). Yet the experience of caring for a child with complex care needs is seen as an evolving process, changing with the illness trajectory and as the child matures (Black, Holditch-Davis and Miles, 2009; Rodriguez, 2009; Murphy, 2016). Parents in this study felt they had no choice other than to deal with the situation they found themselves in, yet when they reflected back on the experience they questioned how they had survived. This study provides evidence that the period following diagnosis or recognition of a life-limiting condition is a challenging time for parents. However, parents felt largely alone and unsupported by service providers at this time. This novel finding could also be attributed to a shift in focus towards home and community-based care for children with complex care needs (DoHC, 2009; Kirk, Glendinning and Callery, 2005). Although the provision of care closer to home for such children is a policy objective, internationally integration of health services is insufficient with wide variation in systems of care for these children and their families (Peter et al., 2011).

In 'Managing an unexpected life' parents endeavoured to respond to the emotional and practical impact of their child's condition on their life and that of their family. While parents strove to achieve normality they equally described their life as not normal. In the context of their expectations of being a parent they perceived their world as different, a world set apart from that of other parents around them. Evidence suggests that there are many factors which differentiate their world from that of the 'real' world. The inherent difficulty associated with accessing help and resources, adherence to schedules, uncertain futures, loss of potential for their child and the fact that their time was never their own is demonstrated both in this study and other literature (Bray et al., 2014; Whiting, 2014). Although the intricacies of what constitutes 'normal' is arguable the sense in this research was many parents felt 'robbed' and as a result, they held on to the illusion of what they thought was normal. For many it was the before and after effect of what their life was like before their child with a NMLLC which affected their perception of normal. However as time progressed the recognition of what life was like now and the dread of what was to come affected their perception of normal. Either way, their focus was on maintaining a sense of normality in their lives. The desire to achieve normality is also highlighted in the literature as a vital component of coping, a process of bracketing out the impact of the child's condition (Price et al., 2011; Steele, 2000; Wood et al., 2010). Deatrick, Knafl and Murphy-Moore (1999) make reference to the 'normalcy lens' suggesting that this is how a family chooses to view their circumstances. According to Paterson's Shifting Perspectives Model of Chronic Illness, living with illness is a complex dialectic between individuals and their world.

Parents in this study often alluded to the daily 'grind' of providing care. Yet at no stage were any explicit complaints made about having to provide this care. Rather, the statements revealed a profound weariness brought about by the difficulty inherent in battling and fighting for help and resources. The findings of a study undertaken by Hunt et al. (2015) with parents of children with life-limiting conditions revealed that they had to fight for and justify their children's and their own needs. Steele and Davies (2006) also propose that despite the negative aspects, that caring for their child was never viewed by parents as a burden. There was, however an undertone of acknowledgement by the parents in this study that their child's condition was profoundly affecting family life. This seemed to be difficult to articulate given their incalculable gratitude that their child had survived often against expectation. In contrast caregiver burden has been described in the literature associated with parents of children with disabilities (Tadema and Vlsakamp, 2010). Jordan and Linden (2013) examined the experiences of mothers caring for a child with an acquired brain injury. The findings provide valuable detail concerning the multi-faceted and perpetual nature of the

burden of caring. Mothers described a ubiquitous anxiety inherent in caring for their child. As a consequence of the quite significant changes in personality, especially those associated with unpredictability in behaviour, mothers ended up at best wary and, at worst, actually frightened by the possibilities of how their child might behave. Furthermore, an awareness of their own progression towards old age and eventual death left mothers particularly fearful for their child's welfare. In contrast, parents in the current study experienced unpredictability associated with their child's health status as opposed to unpredictability in behaviour. Additionally the parents in the current study were aware that their child's life would almost certainly end before their own. In the aforementioned study caregiver burden was, however compounded by a lack of services and support.

In time parents became much more proficient at caregiving. Research with parents of children who are technology-dependent suggests that mastering complex care regimens appeared to develop through experience, resulting in parents developing considerable expertise in the management of their child's condition (Kirk, Glendinning and Callery, 2005). The findings from this research provide evidence to support how over time parents of children with NMLLCs through a combination of bonding, parental knowing, empowerment and acquired expertise take control of the care of their child. Though parents adjustment appeared to be a dynamic process because of the ongoing changes in their child's condition, balanced with varying family needs. The development of parental expertise is described in studies incorporating parents of children with long-term conditions (Smith, Cheater and Bekker, 2015; Swallow et al., 2013). Parents of children with complex chronic health conditions reported mixed feelings regarding the acknowledgement of their expertise by healthcare professionals (Miller et al., 2009). Equally, the parents in this study reported similar sentiments with regard to the recognition of their expertise. Parents often experienced conflicting expectations as they were expected to become experts and assume responsibility for their child's care, but at the same time they were still expected to accept and comply with the advice they received from healthcare professionals. Others felt that their role as expert was recognised by some healthcare professionals, particularly those that were a constant in their lives.

An integral component of the parents experience involved the connections or relationships they had with their partners, family, friends and healthcare professionals. The most important connections described by both mothers and fathers were however those with their partners. Equally mothers and fathers regularly turned to each other in difficult periods. There was a dual benefit in this, in that it helped parents to cope through the difficult periods and also developed a sense of a

supportive connection. Parents of children with chronic illness also reported that shared responsibility with their spouse was a significant source of support (Whittemore et al., 2012). On the contrary experiencing relational disconnect with a partner, which was not uncommon initiated feelings of disappointment, rejection and anger. The negative impact on spousal relationships has also been reported in the literature associated with life-limiting conditions and chronic illness (Kountz-Edwards et al., 2017; Rempel et al., 2012; Smith, Cheater and Bekker, 2015). Bailey Pearce et al. (2017) explored fathers' experiences of their child's life-limiting condition. All fathers acknowledged that their experiences had placed huge demands on the relationship with their partners. Some fathers described how a strong relationship and supporting each other was the key to coping with difficult experiences. Others talked about how the illness had caused arguments as they and their partners, both adjusted differently to the demands and routines of the illness. Interestingly, many of the fathers in the aforementioned study described their initial experiences using the term 'we' as they shared experiences with their partner, suggesting that this is an experience that was co-constructed. However, they did not refer to turning to their partners for emotional support and instead indicated that they felt they needed to be strong and not burden them with their distress. The findings from the current study reiterate the importance of this relationship in the overall experience of caring for a child with a NMLLC.

Previous research has outlined that mothers are usually the primary carer and assume the responsibility of care for children with disabilities and children with complex medical needs (Collins et al., 2016; Green, 2007; Heaton et al., 2005; Toly, Musil and Carl, 2012). In contrast, the majority of the parents in this study were sharing care responsibilities with a partner. This meant taking on an equal share in care arrangements or contributing to the provision of care. This was worked out through a process of negotiation around other commitments, such as work or the need for a break, and was based on a realistic assessment of who was best at achieving the outcome they wanted for their children, irrespective of gender roles. Possible explanations for the difference include that parenting roles are changing and in particular the role of the father has changed to become more integrated in family life. Indeed, in our society, it seems we are facing a slow disappearance of the gender gap and so there should be an equal division of parenting roles within a modern family. Walters (2011) points out that the role of the father has changed dramatically throughout the course of history. In the past gender roles were quite specific, whereas now there is a blurring of gender roles. In addition, Ireland has experienced a trend whereby there are increasing numbers of women seeking paid employment outside of the home (Central Statistics Office, 2016; Office for National Statistics, 2019). There has also been a corresponding increase in the number of men looking after

the home or family. In relatively recent years, Children's health policy has increasingly recognised changing family demographics and roles of fathers in caring for their child. For example, the Children's National Service Framework in England identifies that fathers play an integral role when a child has complex healthcare needs (DoH, 2004).

A small number of studies have indicated fathers' involvement in the care process. The findings of a study undertaken with fathers of children with life-limiting conditions revealed that fathers were engaged in the care of their child, but several felt marginalised by healthcare providers (Nicholas et al., 2016). Loss and adjustment were part of their experience yet they coped in relative isolation and with little support. Additionally, the findings from other studies confirmed that fathers often take up primary caregiver roles or active roles supporting their family with their child's life-limiting condition (Hobson and Noyes, 2011; Wolff et al., 2010; Wolff et al., 2011). Bailey-Pearce et al. (2017) state that when a child has a life-limiting condition, parental involvement is amplified, having to respond to the increased needs of the child. Both parents are affected, yet research has largely under-represented fathers' experiences of their child's condition. Walters (2011) posits that too often healthcare professionals for a variety of reasons fail to engage meaningfully with fathers and that this can result in the absence of vital information that could inform their assessment of and intervention with families. This study provides new insights regarding the active role that fathers assume in the care and management of their child in the context of family life. Importantly, this was expressed through the mothers and fathers accounts. In contrast, the wider literature tends to focus on problematic relationship issues and the high incidence of family breakdown. The inclusion of fathers who are typically less represented in the literature was paramount in this research.

Parents reported feeling guilty and having divided loyalties between caring for the child and meeting the needs of the siblings. In this research, siblings were reported to respond in different ways to having a brother/sister with a NMLLC in their family. Emotional and behavioural changes were common in siblings. Equally, however parents described how siblings became more sensitive and compassionate. Malcom et al. (2014) suggest that siblings are clearly impacted emotionally, pragmatically and relationally by the ill health of another child in the family. Studies have shown that siblings of children with life-limiting conditions present higher levels of emotional and behavioural problems compared with peers (Brennan, Hugh-Jones and Aldridge, 2013; Gan et al., 2017; Read et al., 2010). Other studies have however indicated positive impacts, such as increased maturity, empathy and involvement (Bellin, Kovacs and Sawin, 2008; O' Brien, Duffy and Nicholl, 2009). Interestingly, parents in this study did not refer to sibling support resources. Instead the

findings revealed the importance parents placed on spending time with their well children demonstrated, by the emphasis that the majority placed on setting time aside for them and often using the opportunity respite provided to do so. Respite care was constructed as an opportunity to shift the focus away from the child's everyday care requirements towards the parents' own personal needs and most importantly the needs of the rest of the family. Over time a family balance gained prominence as parents strove to create normality. This included spending time with their well children as well as being able to effectively care for their child at home. Bellin, Kovacs and Sawin, (2008) describe sibling support resources as both limited in number and difficult to access, despite the ways in which peer relations provide a layer of protection for such children.

Parents in this study frequently became isolated from friends, particularly if they were caring long-term for their child. This also triggered feelings of disappointment, rejection and anger. The consequent isolation made the care provided by healthcare professionals all the more important. Isolation has been reported in many studies incorporating parents of children with life-limiting conditions (Collins et al., 2016; Rodriguez, 2009; Somanadhan and Larkin, 2016). Focusing energy back into healthy relationships and caring for their child were common strategies used by parents to distance themselves from disruptive persons. The networks that many parents developed with other parents and families in similar situations formed a vital source of support. Positive interactions with these parents and families made the parents feel they were not alone. These relationships fostered support on physical, emotional and social levels. Social support, particularly an individual's perception of social support is recognised as beneficial (Cohen and Wills, 1985; Sloper, 2000). Akin to other studies, many parents in this study did not have access to a condition specific support group (Jaffe et al., 2010; Pelentsov et al., 2016). In a previously mentioned study with parents of children with life-limiting conditions parents emphasise the importance of living 'ordinary lives' at home (Hunt et al., 2015). This includes taking part in social and leisure activities as well as being able to effectively care for their child. Parents in the current study had, however, very little opportunity for social and leisure activities, particularly if they were caring for other children in the family.

Parents' satisfaction with their relationship with healthcare professionals was variable. The parents were grateful to the few healthcare professionals that went above and beyond their role to care for their child and family and to help them feel supported during times when they felt most vulnerable. Sensitive healthcare professionals were able to enter the life world of parents and understand it from their perspective. Key qualities that parents valued included good communication skills,

empathy, compassion and understanding. Sustained connections with healthcare professionals were important aspects of service provision. However, experiences of relational disconnect with healthcare professionals who did not understand their feelings or vulnerabilities left the parents with powerful memories of hurt and disappointment. Parents perceived that they were unable or unwilling to enter their lifeworld and understand it from their perspective. Bluebond-Langer et al. (2017) suggest that the relationship between parents and healthcare professionals is critical to the care and treatment of children with life-limiting and life-threatening conditions. Maintaining this relationship is central as this will help to avoid a fractured relationship which adversely affects the care of the child and the parents' well-being in survivorship or bereavement.

'The ship is going to go down' exemplified the parents' often never verbalised but ever present experience of living with the knowledge and uncertainty that whilst death was inevitable, they did not know when it was likely to occur. Uncertainty was a common thread interwoven throughout the themes and arguably one of the most difficult aspects of parenting a child with a NMLLC. This finding was also reported in a study undertaken by Sabzevari et al. (2016) which explored mothers' experiences of caring for children with Congenital Heart Disease. Living with uncertainty took its toll on parent's physical, emotional and psychological health. Uncertainty has been acknowledged as one of the most significant and recognised sources of stress for those affected by serious illness (Cohen, 1993; Lin, 2010; Macnamara, 2014). It has been associated with reduced coping mechanisms and a reduction in the quality of life for those affected (Mishel, 1983). Mendes-Castillo, Bousso and Sloand (2017) explored family management in pediatric liver transplantation. The results show that the family's perceived consequences, one of the FMSF dimensions, bring uncertainty about the future as an important issue that deserves attention.

The experience of living with uncertainty has been linked with parents of children with childhood cancer (Molzon et al., 2014; Morrison et al., 2016). The importance of employing coping strategies to reduce uncertainty in families dealing with childhood cancers has also been acknowledged (Gunter, 2018). Though studies undertaken with parents of children with cancer revealed that parents often remembered feeling sure that treatments would be successful to begin with (De Graves and Aranda, 2008; Njuguna et al., 2015). Relapse shattered those feelings, replacing them with a constant sense of uncertainty about their child's survival (Arruda-Colli, Perina and Santos, 2015). In contrast, parents in this study knew that cure was never a possibility for their child and therefore the experience of living with uncertainty was present and pervasive throughout the care trajectory. The rarity of many of the life-limiting conditions and relative lack of healthcare

professional knowledge concerning, for example the illness trajectory intensified the uncertainty experienced. The destabilising effect of prolonged uncertainty has also been studied in families dealing with life-limiting and life-threatening conditions. In research with families dealing with neurodegenerative life-threatening illness, Steele (2000) determined that an overall family goal in the midst of uncertainty was to maintain periods of stability. Similarly O' Brien's (2001) research with parents of children who are technology-dependent at home emphasised the need to attain some degree of stability within an unpredictable situation.

Parents in this study attempted to increase the stability in their lives through a number of coping strategies. These included living one day at a time, using the management behaviour of constant vigilance/hypervigilance regarding the care of their child and through the process of reframing the experience. Parents used these strategies with a reasonable amount of success, although they realised that even these strategies could not totally alleviate the unpredictable nature of family life associated with caring for a child with a NMLLC. Living one day at a time was a universal strategy used by parents to promote family equilibrium. However, parents had multiple concerns about the future, both for their child and for the family as a whole. Steele (2005) described the experience of parents caring for children with neurodegenerative life-limiting conditions as paradoxical in that parents approached life one day at a time and focused on the present and near future but also planned ahead to avoid problems. Parents in this study also articulated concerns regarding the future availability of support and services to assist them to meet the child's ongoing care requirements. Some parents lived in fear that the package of care that they currently had could change for the worse. Correspondingly, the findings of the study undertaken by Hunt et al. (2015) with parents of children with life-limiting conditions revealed that parents expressed fear regarding loss of services resulting from changes in their own circumstances, changes in local policies or reduction in service funding.

Vigilance/hypervigilance was also a strategy used by parents in this study to promote stability in their lives. Examples of vigilant behaviours included constant monitoring of the health status of the child, close attention to the correct and timely performance of physical care and prescribed medications and treatments and careful selection and oversight of all who interacted with their child. These findings are well supported in the descriptions of stories of parents of children with complex care needs and disabilities (Larson and Miller-Bishoff, 2014; Nicholl and Begley, 2012). Parents regarded this vigilance/hypervigilance as a positive element in family life, as it enabled them to foster child and family well-being, ascertain potential problems and intervene in situations before

they became serious and out of control. Likewise Higgs et al. (2016) outlined that parents felt comforted when they could exercise control in maximising their child's quality of life, for example by minimising exposure to respiratory infections and planning special experiences together. However, this constant vigilance was difficult and fatiguing for the parents in this study.

Parents found periods of hospitalisation difficult as they did not have control of their environment. Hospitalisation also exposed parents to many different healthcare professionals, each of whom had time-limited responsibility and interaction with the child and family. Additionally, parents found it difficult to hand over the care of their child to healthcare professionals who, in their perception, often lacked the ability to provide care at the same level as they did. In addition, parents could only benefit from a break when leaving their child in the care of healthcare professionals that they had developed trust and faith in. It has been acknowledged that providing a break for parents of children with complex care needs can only happen if the care of the child is provided by someone who the parents are confident can safely care for their child (McNamara, Dickinson and Byrnes, 2009; Welsh et al., 2014). However some parents expressed concern regarding the competency and experience of professional carers, particularly in the home. 'Questioning competence' likewise arose as a theme in the study undertaken by Brenner et al. (2015). In the said study the majority of parents perceived that many healthcare professionals were unable to care for their child. Moreover a number of parents in the current study expressed dissatisfaction with the inappropriateness of some settings in which care was offered. For example, respite care was occasionally offered in services which were not child specific. As a result, parents regularly rejected these services. The prospect of their child receiving suboptimal care in order that they could have a break was considered unacceptable. Yet the importance of respite for families of children with NMLLCs is widely recognised in research both in Ireland and internationally (Collins et al., 2016; Eaton, 2008; Hunter, 2017; Ling et al., 2016). Wang and Barnard (2008) acknowledged that more respite would considerably reduce the experience of isolation and the feeling of 'being different'.

Finally, a third strategy for promoting stability was that of reframing. Folkman (2001) has stressed the importance of trying to find benefit in a negative experience, arguing that successful coping and positive effect are a result of 'positive reappraisal', a so called cognitive reframing of an experience to be able to view it in a more positive way. Parents in this study adjusted to a new way of living, re-examined their beliefs and values and changed some of their priorities in life. Parents described the joy of parenting a child with a NMLLC. They likewise spoke about the positive benefits and unconditional love the child brings to the family. Since there could be no absolute clarity about how

long the child had to live, parents concentrated on the view that the child had time to live. Parents put even more efforts towards creating a life worth living when their child had limited abilities or when their child deteriorated and lost his/her abilities for life fulfilment. They used coping strategies such as focusing energy on achievable outcomes and looking for the positive qualities in people or situations. Almost all parents articulated a shift in focus from wanting their child to survive to shaping their child's life in a positive way. For parents enabling their child to live a full and active life was extremely important, in part because of their reduced life expectancy, but also to help compensate for the distressing and sometimes painful symptoms children have to live with. Higgs et al. (2016) also outlined a comparable shift in focus in bereaved parents from wanting to cure their child to shaping their child's life and death in a positive way. Research with mothers of children with disabilities supports the importance of reframing, in that it was found to be a significant predictor of positive perceptions of the child (Hastings and Taunt, 2002). Reframing was the most frequently used coping strategy in a study undertaken by Krstić and Oros (2012) with mothers of children with cerebral palsy. Krstić and Oros (2012) described this as encouraging as it helps parents to make their grave situation more acceptable. Undeniably, the experience of caring for a child with a NMLLC had a major influence on the philosophies and worldviews of the parents in this study. The findings had parallels with the work of Corbin and Strauss (1991) which suggest that the management of chronic conditions is more than just a matter of controlling symptoms, or living with disability, or adapting to psychological and social changes that long-term incurable illnesses bring to the lives of the afflicted individuals and their families. It is indeed all of these and more.

The quantitative research findings from the parents provide additional understanding of the unique experiences of parents caring for a child with a NMLLC. These findings reveal that parents' perceptions regarding the care they were receiving were positive. Median scores within each domain of the MPOC-20 indicated that parents perceived that the healthcare professionals' behaviours within each domain occurred to a fairly great extent (median > 5) with the exception of the Providing General Information domain (median=3.5). The MPOC-20 Respectful and Supportive Care subscale received the highest median score in the current study indicating that it is perceived to be the strongest aspect of current service provision. The MPOC-20 Providing General Information subscale received the lowest median score in the current study indicating that it is perceived to be the weakest aspect of current service provision. The findings from this study support the findings of previous research studies which suggest that accessing information is a consistent challenge for parents of children with life-limiting and life-threatening conditions (Collins et al., 2016; Hunt et al., 2015; Menezes, 2010).

Schreiber et al. (2011) undertook a study investigating parents' perspective on rehabilitation services for their children with disabilities using a mixed methods approach. The quantitative and qualitative data relating to parent satisfaction were gathered to cross validate results and offset limitations of using one methodological approach. The findings of their study revealed similar findings to the current study. Median scores within each domain of the MPOC-20 indicated that parents perceived that the professionals' behaviours within each domain also occurred to a fairly great extent (median >5), with the exception of the Providing General Information domain. There was no significant association between the MPOC-20 subscale scores and child age or age at diagnosis in the aforementioned study (Schreiber et al., 2011). Analysis was not undertaken in the current study to investigate if there was a relationship between the MPOC-20 subscale scores and child age or age at diagnosis. In the study undertaken by Schreiber et al. (2011) the subscale responses for Providing Specific Information were significantly associated with receiving more than one service. Higher levels of perception with Providing Specific Information were associated with receiving one service versus multiple services (Mann-Whiney U,  $p < .01$ ). Analysis was not undertaken in the current study to investigate this relationship. However, all parents in the current study were in receipt of care from a variety of settings which could have negatively impacted this subscale score. Similar to previous research, the results of the current study indicate that the provision of General Information and Specific information occurs less frequently than behaviours associated with other domains of the MPOC-20 (Dyke et al., 2006; Hummelinck and Pollock, 2006; Raghavendra et al., 2007).

The findings from this research demonstrate that the MPOC-20 is a useful tool in enabling service providers to identify areas of improvement and move towards services that are more family-centred. Identifying the salient features of parent satisfaction and dissatisfaction with services can aid healthcare professionals and service providers in creating an environment that is truly family-centered (Dickens, Matthews and Thompson, 2011; King, King and Rosenbaum, 2004). Ideally, this will lead to optimal service provision that is responsive to and meets the individual and evolving needs of parents and families as they care for their child throughout the trajectory.

### **6.1.1 Interim comments regarding the unique experiences of parents caring for a child with a NMLLC**

Parenting a child with a NMLLC made a fundamental transformation to the parents' lives. It was difficult, if not impossible for parents to detect an area of their lives in which some personal change had not befallen. Corresponding with Corbin and Strauss's (1991) chronic illness trajectory

framework, the collective efforts of individuals, families and healthcare professionals helped shape the impact of the NMLLC on daily life. The findings suggest that parents in this study used adaptive coping strategies quite frequently while dealing with the situation. Coping appeared to have a bidirectional focus and was aimed at attaining two main goals: ensuring the needs of the child and all family members were met and in accordance with the FMSF maintaining a sense of normality for the child and family. During periods of instability the child's condition remained in the foreground, and at other times, the child's condition remained in the background. In keeping with Paterson's (2001) Shifting Perspectives Model of Chronic Illness when the child's condition was in the foreground, the focus was on the illness, but when it was in the background, the focus shifted to the health and well-being of the entire family. This highlights the importance of perception and helps to explain how families develop the normalcy lens, an attribute of normalisation. Consistent with previous research the findings demonstrate that service provision for children with NMLLCs has impact far beyond the child and the care they receive. It impacts the entire family. Thus services should aim to support family functioning. Even though parents reported some positive aspects of current services, for example their relationships with some healthcare professionals and the quality of certain services they received the overall research findings highlighted a lack of available services, concern regarding the quality of certain services and difficulties associated with accessing services. Further discussion with regard to these findings will be undertaken later in the chapter. Parents' views of accessing services when caring for their child will be discussed next.

## **6.2 Parents views of accessing services when caring for their child**

Bronfenbrenner (1979) argues that due regard should be given to other environmental influences such as the system that impact on both the child and family therein. This section provides an understanding of parents' views of accessing services when caring for their child. It was apparent that the experience of caring for a child with a NMLLC was overwhelming in terms of finding and accessing services and support. The findings reveal that parents had to fight for services and support in order to manage to care for their child. In their fight parents regularly encountered inconsistent and inequitable services often characterised by bureaucracy and delay. This was true irrespective of the nature of the service required. Bureaucratic delay in the provision of essential services compromised the care for the whole family. In addition services were not necessarily sufficient or indeed sensitive to the needs of the child or the family. A lack of uniformity in all aspects of service provision and service quality was also highlighted. Moreover, there was a sense in which the very act of seeking /securing support, something which should assist parents actually served to intensify their physical and emotional weariness. Hunt et al. (2015) state that whilst parents often come to

view themselves as 'experts' in the care of their child, the availability of appropriate support is identified as an essential resource for families of children with life-limiting conditions.

Dwybik et al. (2011) explored the experiences of families giving advanced care to family members (children and adults) dependent on home mechanical ventilation. The core category, 'fighting the system,' became the central theme as family members were asked to describe their experiences. All of the participants expressed a strong desire for their ventilator-dependent family members to be given the opportunity to live the most optimal and normal life as possible despite serious respiratory failure and severe disabilities. Despite considerable effort and commitment, the families were still dependent on their local community health care services to ensure their loved one received proper care. Based on the extensive experience the participants had, they were mostly focused on describing the continual struggle with the community health care services, or 'the system', as they called it. Even though a member of their family was completely dependent on highly advanced medical procedures and technological equipment, the 'fight against the system' seemed to be the most problematic issue. 'Fighting and frustration' emerged as a theme from the interviews which explored parents' perceptions of the transition to home when a child has complex technological healthcare needs (Brenner et al., 2015). Each of the parents interviewed expressed their anger and frustration at the length of time it took to have everything in place so they could take their child home and how hard they had to fight to secure and retain homecare for their child. Parents of children with disabilities also highlighted the need to fight for their child and the care they received (Bye and Aston, 2016; Todd and Jones, 2005). Parents of children with life-limiting conditions likewise spoke of 'battling' for services and entitlements (DoHC/IHF, 2005).

Other studies in Ireland provide valuable information regarding service provision. Price et al. (2012) examined the experiences of bereaved parents concerning the care provided to children who died from cancer to those who died from a non-malignant life-limiting condition in Northern Ireland. Although parents' accounts displayed commonalities, key differences were discernible. Typically, parents of children with cancer considered care at end-of-life as well-resourced and responsive to their child's needs. In contrast, parents of children with non-malignant conditions reported under resourced and inadequately responsive services. Parents of children with non-malignant conditions where cure was never a possibility described expending huge amounts of emotional and physical energy in their battle for adequate service provision, aware of having to struggle on what they saw as two related fronts: bureaucratic ineptitude and inadequate resources. McVeigh et al. (2018) explored the experience of palliative care service provision for people with non-malignant

respiratory disease and their family carers in rural and urban areas in the North and Republic of Ireland. Even though this study was undertaken with adults commonalities between this and the current study were apparent. Inequalities in the provision of palliative care services across the island of Ireland were evidenced in both studies.

Accessing respite care was particularly problematic for parents in this study. The parents encountered multiple obstacles with regard to access resulting in stress, confusion and frustration. Parents were often left to reach breaking point, experience ill-health and lead diminished lives as they struggled to care for their child. While respite care was viewed by most as essential this did not preclude from parents feeling guilty when they were granted it and started using it. For parents who had access to out-of-home respite services they sought adequate staffing, well maintained facilities and for their child to have an enjoyable and stimulating time while at respite. Neufeld et al. (2001) suggest several reasons why parents may be reluctant to use respite services. These include guilt and concern about leaving the child with a stranger, loss of privacy and previous experience. Parents in the current study often reported that the real value of respite was the opportunity it afforded to spend time with their other children. However, they also spoke of the need for a break as a central motivation for accessing respite care. Yet the time and effort required to circumvent the numerous barriers encountered often influenced parents overall perception of the extent to which they actually obtained 'respite' from respite care. Respite care provision was generally described as inadequate. In a review of palliative care services for children and young people in England Craft and Killen (2007) also identified access to respite care as a key problem suggesting that there were varying acceptance criteria among providers. Moreover, Thomas and Price (2012) undertook a study to evaluate the experience of nursing respite care in families caring for children with complex healthcare needs. The findings reveal inconsistency in the amount and types of respite provision offered to families and indicate that fewer respite opportunities are available to the family when the child deteriorates.

The findings from the demographic questionnaire provide additional information regarding the issue of accessing services. The findings reveal that all parents had access to in-home respite care. A small number of parents were not utilising the in-home respite that was available. In contrast, out-of-home respite was not available for all parents to utilise. The most remarkable finding was that some parents had access to two out-of-home respite services whereas others simply did not have access to any. Again for those parents who had access to out-of-home respite care not all were utilising it. Ling et al. (2015) outlined that parental decision-making regarding the utilisation of out-

of-home respite is dependent on many interlinking factors including the child's age, diagnosis, geographical location and the family's capacity to meet their child's care needs. Additionally, in order for parents of children with life-limiting conditions to consider out-of-home respite it is essential that the respite offered to them is provided in an acceptable form and location. Urban dwellers had better access to suitable out-of-home respite care than those living in rural regions. The findings of this study confirm that the provision of out-of-home respite services to children with NMLLCs in Ireland is still dependent on geographical location rather than need (DoHC, 2005; IHF/LLH, 2013). Whiting (2014) discovered the greatest area of unmet needs identified by parents is the provision of respite care.

Navigating appointments, particularly in the time period following diagnosis or recognition of a life-limiting condition took up a great deal of time in addition to the daily care giving duties, creating added challenges for parents. All journeys required careful planning and pre-emptive care. For many parents the frequency of appointments decreased in time. Additionally, parents described efforts that were made to structure appointments to ensure a coherent approach to care. Nicholl (2015) explored mothers' experiences of caring for a child with complex needs at home in Ireland. The findings suggest that the delivery of care to children with complex needs involves the provision of care in three different environments, in the home (the inside world), outside the home (the outside world) and in travel the going between world. Mothers were required to attend multiple appointments with their children. They had to attend local and national centres to meet their child's needs. Few mothers reported on any attempt to coordinate services to prevent multiple visits. Care provision while travelling was challenging. Significant travel preparations were meticulously planned and every eventuality was thought out. As a consequence few unnecessary journeys were undertaken. The findings in the current study are similar in that parents also indicated that travelling with their child was difficult. The parents suggested that they could not immediately leave home because they needed additional time to schedule and prepare than 'normal' families. However, in contrast, parents in this study reported efforts to coordinate services to prevent multiple visits.

In some respects, some positive changes were gradually being evidenced over recent years by some parents in terms of services available. This change is most likely due to the growing awareness and understanding of CPC (DoHC, 2009; Hain, Heckford and McCulloch, 2012). Most parents had access to a Children's Outreach Nurse/ Link nurse or nurse specialist who assumed the role of key worker. The allocation of a key worker has been highlighted in the literature as valuable (Greco et al., 2006;

Hillis et al, 2016). It is suggested that they can navigate the system on behalf of the families and can ensure that families are adequately supported. Hsiao et al. (2007) highlight the strength of having access to a healthcare professional quickly and easily and receiving a prompt response. For parents who had access to a team of caregivers, being able to access members of the team easily and all on one day was greatly appreciated. In the previously mentioned review of services in England Craft and Killen (2007) also revealed geographical variation across and within the different regions, with some examples of excellent practice, yet many areas in which service provision was inadequate. The findings of the current study argue strongly for the importance of a key worker and a team of healthcare professionals who oversees and coordinates the care of each child.

A report undertaken by the Irish Hospice Foundation (IHF, 2016) found that significant progress has been made on the implementation of key recommendations contained in the National Policy- Palliative Care for Children with Life-limiting Conditions in Ireland (DoHC, 2009). The feedback from parents indicate that the introduction of the Children's Palliative Care Programme improved the quality of life of children and parents in a wide range of ways including home-hospital-home transition, securing resources and equipment for children at home, providing expert advice on symptom management and helping parents in their role as primary caregivers in the home. Parents identified ongoing support needs which include access to quality and consistent nursing hours, availability of respite and improved access to equipment and supplies. The valuable role of the Children's Outreach Nurses was a recurring theme. Having one 'go-to' person was consistently described by parents as greatly easing the burden of care and improving continuity of care for children and families.

The findings suggest that the need for quality services is high (in particular respite care) yet access is limited. In general, advances in medical healthcare that enable children with NMLLCs to survive for longer are not mirrored with advances in supportive health care systems. Not all parents chose to take advantage of the services offered, but knowing that they were there meant that they could continue to care for their child. Price et al. (2011) explored bereaved parents' experience of living through the death of a child. The findings indicate that even having the contact details of relevant healthcare professionals provided parents with reassurance and (anticipated) support irrespective of actual use.

The findings from the MPOC-20 reveal both convergence and discrepancy or dissonance to the qualitative research findings from the parents. The results from the MPOC-20 demonstrate that parents' perceptions regarding the care they were receiving were positive. Yet the qualitative research findings from the parents reveal dissatisfaction regarding many aspects of care provision. However, parents talked about the quality of certain services they were receiving. Additionally, parents were receiving services from a variety of service providers yet the focus when completing the MPOC-20 was on one service provider. The results demonstrate that the strongest aspect of service provision (as indicated by the highest median score) is related to the type of interaction and relationship parents have with healthcare professionals rather than any practical aspect of service provision. Yet parents' satisfaction with their relationship with healthcare professionals was extremely variable. The MPOC-20 domain Coordinated and Comprehensive Care were ranked high. Yet the parents indicated that care provision particularly at the time of diagnosis or recognition of a life-limiting condition was uncoordinated and unstructured. However, this aspect of care appeared to improve in time and in particular areas. Additionally the scores on this particular domain ranged considerably, reflecting different views with regard to whether the care received was in fact coordinated and comprehensive.

The MPOC-20 domains Providing General Information and Providing Specific Information were ranked lower than the other domains for parent satisfaction and correspond with qualitative data. It is clear from this study and previous research that information provision is necessary for a basis of family-centred care (Hummelinck and Pollock, 2006). One of the most pressing issues for parents was the lack of comprehensive information available to help them avail of potentially useful and even essential services. Parents frequently referred to the process of gaining useful information as 'haphazard'. They suggested that information provision must be tailored to the individual needs of the parents and families. This study and other studies suggest that information provision beyond the patient's needs and preferences may increase mental strain and anxiety and reduce confidence (Fisher, 2001; Hummelinck and Pollock, 2006). It is therefore critical to identify and re-evaluate on an ongoing basis parents' needs for information, to provide that information in a usable format and to monitor parent understanding of the information (Hummelinck and Pollock, 2006). The provision of information was also essential for decision-making which will be discussed next.

### **6.3 Decision-making regarding the care and management of children with NMLLCs**

This section provides an insight into decision-making regarding the care and management of children with NMLLCs from the perspective of parents. Parental decision-making commenced at the time of diagnosis or recognition of a life-limiting condition and continued throughout the care trajectory. Evidence similarly suggests that parental decision-making can span the care trajectory from diagnosis of a life-limiting condition through to end-of-life care (De Vos et al., 2015; Ling et al., 2016; Rodriguez and King, 2014; Tamburro et al., 2011). Bluebond-Langer et al. (2017) put forward that as treatment options increase so do choices and decisions that need to be made. Beecham et al. (2017) propose that decisions may encompass the initiation, continuation or withdrawal of therapy including ventilation, changes in places of care and approach to nutrition. Parents in this study encountered an array of challenging decisions as they commenced on their care journey.

As parents were 'Starting out in haziness' they entered an uncharted world of decision-making. Through this emotionally difficult time period, parents were confronted with numerous challenging decisions and considerable uncertainty regarding the impact of their decisions on themselves and their families. Toebbe et al. (2013) also revealed that parents of infants with Hypoplastic Left Heart syndrome encountered life or death decisions within days of their infant's birth. In their study, they outline the importance of parental support for early decision-making. Decision-making continued for this cohort of parents throughout the child's life with each prior decision affecting the next decision. Through this time period, parents in the current study were often required to make decisions in a dynamic and time-sensitive manner. Their powerful emotions, the care environment and the nature of the decisions affected information processing and their ability to make decisions. Other studies have reported that parents of children with life-threatening conditions likewise struggled to understand and integrate the illness and treatment options after receiving the diagnosis (Boss et al., 2008; Rankin et al., 2016). Consequently, parents in this and other studies frequently made decisions centred on the child's survival (Boss et al., 2008). The findings of a study undertaken by Payot et al. (2007) with parents of extremely premature infants suggest that parents are certainly able to grasp the information they receive but tend to reformulate its significance in terms of the possibility of a positive outcome in order to counterbalance the uncertainty and pessimism of the multiple risks that are conveyed to them.

Alternatively, parents handed over decision-making responsibilities to healthcare professionals. Beecham et al. (2017) propose that there are times when parents are happy to go along with the recommendations given by healthcare professionals. Furthermore, the parents' role in decision-making at this time was often limited due to the urgency of the situation and the seriousness of the condition. Parents were often aware that they were giving permission as opposed to making an active decision as they needed to do what the healthcare professionals recommended. A number of parents regretted the decisions that they made at this time as they proceeded on their care journey. This is a significant finding as parental regret regarding decision-making can lead to psychological distress, decreased physical health and decreased quality of life for parents (Brehaut et al., 2003; Korenromp et al., 2005). Bluebond-Langer et al. (2017) propose that depending on the decision taken, harm may be done and suffering may result. Parents must live with the decisions that they have made. Trowbridge et al. (2017) propose that decision-making is often conceptualised as binary, in which parents are asked to choose therapeutic or palliative approaches at one time point. However, in most instances and as evidenced in this study decision-making is both dynamic and evolutionary. Additionally decision-making did not occur in isolation rather parents decision-making was influenced often in inconsistent ways by a diversity of factors. For example, the health status of the child had variable effects on parents' decision-making.

In 'Managing an unexpected life' parents became more familiar with this world of decision-making and were largely clearer in their thinking. Whereas in the time period from diagnosis or recognition of a life-limiting condition they experienced difficulties in understanding and retaining complex explanations regarding their child's condition they now felt empowered in decision-making through the provision of accurate information in a sensitive and timely manner. Parents also recognised the limits of individual healthcare professional's knowledge and found in some cases their own decision-making was hindered by healthcare professional's inadequate condition-specific knowledge. There appeared to be a conflict between parent's needs and desires for information and the providers abilities related to offering information. Bluebond-Langer et al. (2007) revealed that parents sought and/or relied on healthcare professionals' knowledge about which treatment options were best for the child when standard cancer treatment had failed. Studies with parents of high risk premature infants also relied on information to make decisions throughout the child's life (Grobman et al., 2010; Moro et al., 2011).

The type of decisions parents faced often changed at this time and parents regularly had more time to contemplate the decisions. Choices were still not easy for parents but were necessary in order to manage. Decisions were made at this time in the context of both the child and family. For example, some parents made a decision to avoid certain settings or situations to minimise infection for the child. At other junctures, parents consciously chose not to lay emphasis on the life-limiting condition and accepted the risk of deterioration in favour of a good family balance. For many of the parents the decision to accept help was influenced by the accessibility of the services, trust in service providers and the needs of the family. This study showed that it is not only the complex palliative care and end-of-life decisions but also the numerous minor decisions related to daily life that required attention and efforts from parents. However CPC research mainly focuses on end-of-life decisions (Carnevale et al., 2006; deVos et al., 2015; Hinds et al., 2009). Owing to the unpredictable nature of NMLLCs the need to contemplate challenging decisions was always near. These decisions once again regularly provoked strong emotions which on occasion were complicated by misunderstandings between parents and healthcare professionals. Likewise Boss (2008) revealed that parents once again struggle to understand and integrate the illness and treatment options during other critical changes within the illness course. Parents indicated that an understanding of the situation and an appreciation of what to expect was imperative in preparing them for decision-making.

Parents spoke about how it was essential to contemplate decisions regarding their child's future knowing that 'the ship is going to go down'. However, making decisions about future treatment options was difficult because treatment options were hypothetical, and their preferences might change in the future as circumstances changed. At the heart of decision-making was the desire to be good parents. Verberne et al. (2017) explored parental caregiving based on the lived experience of parents caring for a child with a life-limiting condition at home. These parents expressed their desire to be a good parent for their child in the extraordinary situation of knowing that their child's life is limited and within an unknown time span. They strived for three main aims in caring for their child. Firstly, they primarily aimed for optimal controlled symptoms and controlled disease. Over time a life worth living and a family balance gained importance. Since the time with their child was limited, parents developed a major need to concurrently achieve each separate aim and felt under pressure because everything had to be as good as possible. However they could not always succeed herein, resulting in considerable distress for parents. Key to understanding decision-making is the roles which parents and healthcare professionals occupy in their lives (Bluebond-Langer, 1980; Bluebond-Langer, 2000; Bluebond-Langer et al., 2007). A role is something constitutive of one's identity and

sense of self (Bluebond-Langer et al., 2017). It significantly affects how we conduct ourselves and how we feel about those interactions. Part of the role of a parent is to protect and advocate for their child. As evidenced in this study this begins at the time of diagnosis or recognition of a life-limiting condition and continues throughout the care journey. Bluebond-Langer et al. (2017) propose that advocating can take the form of dealing with options for treatment. Protecting involves weighing the risks which they present. Driven by the need to leave no stone unturned, parents' advocacy is tireless.

Decision-making was also guided by what parents perceived to be in the best interest of the child and quality of life considerations. Michelson et al. (2009) explored parental views on withdrawing life-sustaining therapies in critically ill children. Parents described aspects of quality of life that might influence them to consider withdrawing life-sustaining therapies. Some parents simply used the term 'poor quality of life'. Others described suffering neurological dysfunction and many described scenarios in which their child could not interact with his/her surroundings. Dependence on machines was also discussed frequently. However, most of the children in the said study did not have a life-limiting condition. Thus the findings may be difficult to compare with the current study where the majority of the children had neurological dysfunction associated with their life-limiting condition. Although parents described how they were guided by what they perceived to be in the best interest of the child, they also described feeling conflicted on occasion. They described being torn between what they thought was in the best interest of their child and what they 'as parents' wanted. Beecham et al. (2017) described similar sentiments in a study which investigated how parents of children with life-limiting conditions approach and experience advance care planning. Parents expressed conflicted feelings about these decisions because whereas they didn't want their child to suffer, they also wanted to do everything possible to try to increase the length of their child's life. In addition decisions were often influenced by anticipation of regret. Parents did not want to live the rest of their lives with the knowledge that it had been their decision to pursue no further treatment because for them that carried the implication that they had 'given up' on their child. Much psychological research on regret has been based on the premise that it is a functional emotion that help shapes good decision-making (Camille et al., 2004; Zellenberg and Pieters, 2007).

The parental role in decision-making has changed over the past decade giving parents more autonomy about how they personally believe their child should be treated (Allen, 2014). Oshima and Emanuel (2013) propose that shared decision-making has particular utility for treatment options which do not always have a single superior option because it incorporates values to make an optimal

decision for the individual. Although parents' preferences for degree of participation in decision-making varied to begin with, as time progressed most were interested in sharing decisions with trusted healthcare professionals. Previous research with parents of children with special healthcare needs found that parents exhibited different attitudes towards involvement in decision-making: some wished and expected to be involved, others preferred not to be involved (Zanello et al., 2015). Rodriguez and King (2014) reported that parents had a desire to be involved in care decisions but did not necessarily want to make the final decisions. Wright et al. (2009) recognise that some parents will always want to be active decision makers whereas others will want to defer decisions or ask healthcare professionals to make choices for them. While parents in this and previous studies indicated a preference to be involved in discussions and decisions, their chosen level of involvement was influenced by timing with regard to illness progression and the nature of the decision (Caeymaex et al., 2013; Sullivan, Gillam and Monagle, 2015). Parents acknowledged the importance of healthcare professional support with regard to decision-making.

Allen (2014) state that regardless of the type of decision, parents work with healthcare professionals to determine the optimal choice for the child. All parents prominently mentioned the interaction between themselves and the healthcare professionals, including the need for healthcare professionals to understand the bigger picture of the life of the child and the life of the wider family rather than simply focusing on treating a particular symptom. Parents in this study reported positive relationships with healthcare professionals and healthcare professionals' ability to listen to them as experts in the care of their child. When communication between parents and healthcare professionals was efficient, parents experienced less anxiety and became more involved in decision-making which in turn encouraged their autonomy. Trowbridge et al. (2017) also highlight the importance of allowing parents time given the complexity inherent in the decisions they encounter. However, parents also recounted when they were not listened to or not sufficiently involved in decision-making and this created a great deal of stress. The insensitive manner by which some parents were approached, the limited information they received and the pressure that was exercised upon them to reach a decision contributed to their refusal to engage with healthcare professionals. The approach of avoiding discussions with healthcare professionals was viewed as protective by parents. Wright et al. (2009) state that it should not be taken for granted that the perceptions of families mirror those of healthcare professionals. De Vos et al. (2015) concluded that parents are able to handle a more active role than they are currently being given regarding end-of-life decisions.

Other key factors influencing parental decision-making included hope, religion and spirituality. Parents reported that they could not trust healthcare professionals who showed no hope. They particularly doubted those few healthcare professionals who told them with conviction that their child would not have a good outcome. In a study undertaken by Kavanaugh, Moro and Savage (2010) the lack of hopeful communication led to a strained relationship between the parents of extremely premature infants and healthcare professionals. Boss et al. (2008) also revealed that parents needed to feel that healthcare professionals were compassionate and hopeful as these behaviours demonstrated the healthcare professionals respected their child as an individual. Religious and spiritual beliefs were important to parents in this and other studies when making decisions about withholding or withdrawing treatments (Ahmed et al., 2006; Michelson et al., 2009). Some parents relied upon God or faith to guide their decision-making (Sharman, Meert and Sarnaik, 2005). Others felt that the decision was up to God and not to be made by humans (Pepper et al., 2012).

Parents reported that when making decisions about their child's care and treatment, they had considered several factors: some were focused on the ill child or family as a whole, others focused on reasons of their own or on advice given by healthcare professionals. This study identified several ways to support parents in making decisions. Examples include, as also reported by parents in other studies, the sharing of information by trusted healthcare professionals, information about the potential outcomes of treatments and the consequences from refraining from certain options (Dellon et al., 2010; Sullivan, Monagle and Gillam, 2014).

Légaré et al. (2012) suggest that decisional conflict is a central determinant of decision-making particularly in the context of uncertainty. As outlined in the qualitative research findings parents in this study regularly faced decision-making dilemmas. Decisional conflict was measured using the Decisional Conflict Scale (DCS) (O' Connor, 1993). The findings provide an enhanced understanding of decision-making. Parents in this study reported a generally low level of decisional conflict in the DCS total scale and subscales. The median score on the DCS total scale was 27. As described in the instrument developer's manual, a score of 0 indicates low decisional conflict; a score of 25 is associated with implementing decisions and scores of 37.5 and higher are associated with decision delay or uncertainty about making a decision (O' Connor, 1993). Geurtzen et al. (2017) evaluated decisional conflict and regret among parents in initiating comfort or active care in extreme prematurity. The median score on the DCS total scale was 28. Overall the findings similarly revealed little decisional conflict and no decision regret among parents regarding decision-making at twenty-

four weeks gestation. From the subscales within the DCS, the DCS Uncertainty subscale revealed the highest median score of 42 in the current study-revealing that parents felt uncertain about best choice. In the aforementioned study the DCS Values Clarity subscale revealed the highest median score of 42-revealing that parents felt most unclear about personal values for benefits and risks of the decision on either comfort or active measures (Geurtzen et al., 2017). The DCS Effective Decision subscale received the lowest median score in the current study. This is encouraging as the findings of a study undertaken by Knapp et al. (2014) with parents of children with life-threatening conditions reveal that scores on the DCS Effective Decision subscale were significantly lower among parents reporting a more positive impact of the child's illness on the family.

In the previously mentioned study Knapp et al. (2014) sought to investigate how decisional conflict varies among racially and ethnically diverse families of children with life-threatening conditions. Results from the unadjusted bivariate analyses demonstrate that minority individuals would have greater decisional conflict than Whites. Knapp et al. (2014) propose that conflict in decision-making among minorities, especially in cases of life-limiting conditions may often be related to mistrust among physicians and the medical community. The sample in the current study was, however homogenous in terms of ethnic profile with all parents of Irish ethnic background. Thus, this type of analysis could not be undertaken. However the fact that the sample was homogenous in terms of ethnic profile could be a contributing factor to the generally low level of decisional conflict experienced by parents in this study. Knapp et al. (2014) also found that decisional conflict was related to whether a child had a life-threatening condition at birth or whether they developed a life-threatening condition later in life. The DCS Values Clarity subscale for example, revealed lower scores for children who were diagnosed with their life-threatening condition at birth as compared to children diagnosed at a later time. Analysis was not undertaken in the current study to investigate if there was a relationship between the timing of diagnosis and DCS scores. Knapp et al. (2014) also found that single-parent households had more conflict compared to two-parent households across one domain. They suggest that being able to talk through decisions with someone else provides decision-makers with a different perspective. The majority of parents in the current study were two-parent families which could account for the generally low level scores on the DCS Support subscale.

Importantly the scores on the DCS total scale and subscales indicate that a small cohort of parents experienced high levels of decisional conflict in the current study. This is noteworthy as decisional conflict can be lowered with decision supporting interventions (O' Connor, 1993; Oshima and Emanuel, 2013). Information about options, benefits, risks and side effects can make parents feel

more informed. Values can be clarified using strategies such as: describing outcomes in sufficient detail to better judge their value. Parents may feel more supported in decision-making if they are guided in the steps of deliberation and shared decision-making. As a consequence, their uncertainty stemming from these modifiable factors may decline and they may feel they have made a better decision. By better decision, they may feel that they have made a more informed value-based decision, are more likely to stick with their choice, and are more satisfied with the decision. Chiavari et al. (2015) undertook a pilot study to evaluate the effects of a decision-making support (decision counselling) on decision-making, decisional conflict and anxiety in cancer patients facing values-based decisions on fertility and procreation or genetic testing and risk reduction options in oncology. The findings demonstrate that decision counselling is useful to facilitate and reduce decisional conflict. This confirms previous research findings that report the efficacy of decision support in reducing decisional conflict and promoting awareness with value-sensitive choices (Stacey et al., 2011).

Parents were asked about decision-making in general as opposed to specific decisions such as the decision to limit life-sustaining treatments. Specifying the decision might yield different results for this cohort of parents. The DCS is an important tool for researchers and healthcare providers who are working with parents who are deciding whether certain decisions are the right choice. As indicated in this study the DCS can identify parents who experience high decisional conflict, so they can receive more clinical attention and decisional support. The development and use of decision support tools should be explored in parents of children with NMLLCs. The ability to measure decisional conflict in a valid and reliable manner should provide useful information that can be used with evidence-based interventions to support parents in decision-making and reduce decisional conflict (Knapp et al., 2010).

The findings from the DCS reveal convergence and discrepancy or dissonance to the qualitative findings. Parents in this study reported a generally low level of decisional conflict in the DCS total scale and subscales. Yet a number of parents regretted the decisions they made at the time of diagnosis as they proceeded on their care journey. However the scores on the DCS total scale and subscales indicate that a small cohort of parents experienced high levels of decisional conflict in the current study. From the subscales within the DCS, the DCS Uncertainty subscale revealed the highest median score of 42 -revealing that parents felt uncertain about best choice. Parents also spoke about how they experienced uncertainty with regard to decision-making. They were guided by what they perceived to be in the best interest of the child but also described feeling conflicted on

occasion. They described being torn between what they thought was in the best interest of their child and what they 'as parents' wanted. The DCS Effective Decision subscale received the lowest median score in the current study. The findings of a study undertaken by Knapp et al. (2014) with parents of children with life-threatening conditions reveal that scores on the DCS Effective Decision subscale were significantly lower among parents reporting a more positive impact of the child's illness on the family.

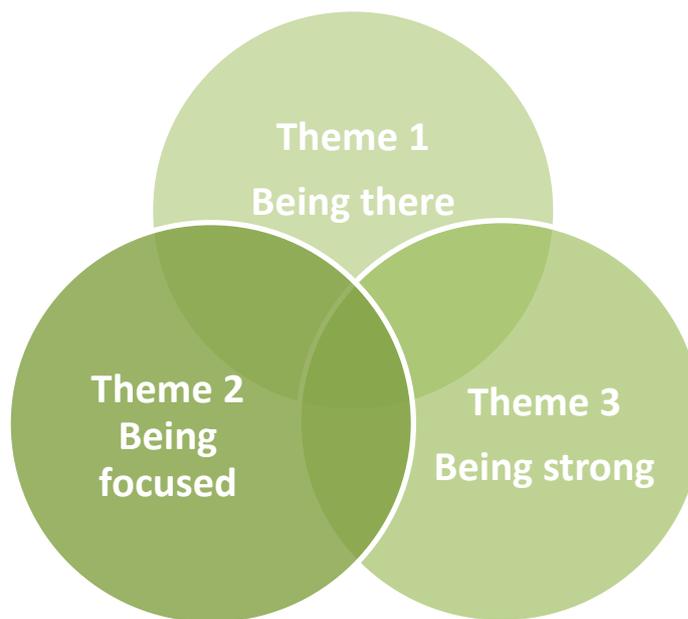
## **6.4 Concluding comments**

This chapter has presented a detailed discussion of the qualitative and quantitative research findings from the parents with reference to the initial study aims and objectives and set in the context of the current evidence base. This included an integrative discussion. Parents in this study identified a range of experiences in the complex nature of their caring activities. They outlined the overwhelming nature of their experience as they commenced on their care journey. Within the literature parents of children with life-limiting conditions described the initial experience as traumatic, overwhelming and confusing (Collins et al., 2016; Rempel et al., 2012; Somanadhan and Larkin, 2016). There appeared to be a less significant focus in the existing literature regarding the professional support parents received in relation to their role as caregiver a topic area which this study has addressed. The current study clearly identified the lack of professional support for parents in relation to their role as caregiver. The findings from this research corroborate with previous research which suggests that decision-making regarding care and interventions for children with NMLLCs is fraught with difficulties but faced regularly by parents. Parents in this study reported a generally low level of decisional conflict. However, the findings indicate that a small cohort of parents experienced high levels of decisional conflict. This was interpreted as noteworthy as decisional conflict can be lowered with decision supporting interventions (O' Connor, 1993; Oshima and Emanuel, 2013). Parents reported a generally positive perception of family-centred care provided. The next chapter will present the qualitative research findings from the healthcare professionals.

## Chapter 7: Presentation of findings: Healthcare professionals

### 7.0 Introduction

This chapter presents the findings from the analysis of the twelve healthcare professional interviews. As indicated previously, Braun and Clarke's (2006) six-phase framework for thematic analysis was used to help guide the analysis of the data. This resulted in three dominant themes and eight subthemes emerging, charting the healthcare professionals' experiences of caring for children with NMLLCs, including their experience of service provision and decision-making. The three dominant themes are diagrammatically represented in Figure 9. The relationship between the three dominant themes are then discussed. Following this, each theme is elucidated and the subthemes explained. Explicit discussion of their relevance or how they answer the research objectives will be undertaken subsequently.



**Figure 9: Diagrammatic representation of the themes: healthcare professionals**

## 7.1 Relationship between the themes

During ongoing thematic analysis of the interviews it was apparent that a relationship existed between the three themes. All three themes were intrinsically interlinked. Theme 1 'Being there' explicitly highlighted expert care that was individualised and responsive to the unique needs of children with NMLLCs and their parents and family throughout their care trajectory. The provision of such expert care was often threatened and potentially compromised by the intricacies and challenges associated with CPC and service provision. Theme 2 'Being focused' captured the sense that the healthcare professionals remained entirely focused on providing expert care despite these challenges and intricacies. Both 'Being there' and 'Being focused' impacted both personally and professionally on the healthcare professionals. Theme 3 'Being strong' epitomised the personal and professional impact on healthcare professionals who are working in the area of CPC and the manner in which they sustain themselves in 'being focused' and in their 'being'. 'Being strong' without doubt enabled the healthcare professionals to 'be there' and 'be focused'. The relationship between the themes is also evident in Figure 9. For the purpose of clarity the three themes are presented separately.

## 7.2 Being there

This analytically derived theme explicitly highlighted expert care that was individualised and responsive to the unique needs of children with NMLLCs and their parents and family throughout their care trajectory. 'Being there' captured a sense of determination, dedication, passion and empathy on the part of the healthcare professionals and appeared borne from their knowledge that families needed support amidst emotional and practical chaos. The healthcare professionals repeatedly spoke of the significance of 'being there' with many being present throughout the care trajectory from diagnosis through to end-of-life care and following the death of the child:

*I find that being there is very important. I mean, I think sometimes it's not about sorting it out, it's about being there as a presence, as a listening ear, as a support and normalising what parents are going through at any one time. [P11].*

The healthcare professionals revealed the true value of 'being' as opposed to always 'doing'. They realised that their duty was not to solve every problem or complex situation but to 'be there' to support parents and families.

'Being there' was, however incredibly complex and multi-faceted. A component of being there for the healthcare professionals involved fully embracing their role of advocate in an effort to support children and their parents and family:

*We are very much an advocate for that family, not just within the hospital and within the multidisciplinary team that we work with. We are their advocate both in the hospital and in the community. We need to advocate for them. [P11].*

As advocates, the healthcare professionals made every effort to ensure that the families' needs were met and that their best interests were respected. 'Being there' was therefore much more than just being there from a physical perspective; it incorporated many different facets:

*It's much more than just talking. I could be in a house listening, talking ...directing. And there is so much more going on than would appear. There is so much more happening and so much going on. [P3].*

In 'being there' the healthcare professionals frequently assumed numerous roles to varying degrees, including healthcare provider, case manager, educator and counsellor throughout the course of the care trajectory.

The close relationship that healthcare professionals developed with children, parents and their families were viewed as key to 'being there'. This was true irrespective of duration that they had to develop this relationship:

*I think such a big part of it is getting to know the families, which is the bit that's very hard to measure. And yet I think it's the core. That's the bit that makes the difference, for me....yourself as a resource. [P3].*

Certainly, caring for a child for a long period assisted healthcare professionals in getting to know the child which formed the basis of this relationship. It also helped them get to know the parents' personalities which meant they had a better understanding of how to support them: "you've got to know where they're coming from that's really, really important." [P12]. In addition, trust was viewed as critical to the relationship. This relationship did not happen by chance but rather required a conscious effort on the part of the healthcare professionals' in establishing and maintaining it. In some situations, there was limited time to develop a relationship with parents and families due to the nature of NMLLCs in children and the pattern in which they presented:

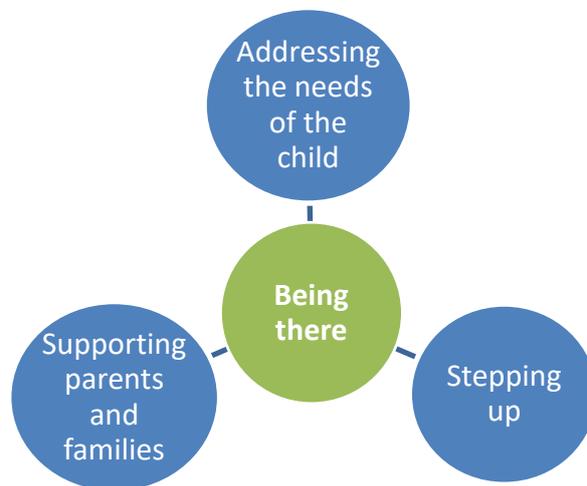
*I recently had that issue, where I was referred a baby... now, in fairness, the baby was only six months old, and she was referred three weeks prior to her passing. So that was a challenge in that I was clearly coming in where this baby was having worsening seizures and the neurologists were saying there's no more drugs to try. And that was hard, because I was giving that information, even though I didn't know them. Why should the parents trust you when you've only seen their baby for a few days? That does happen. That's not the norm, but that can happen. [P4].*

It was challenging for healthcare professionals to develop a rapport and establish trust over shorter periods. However, the healthcare professionals were experienced and had developed expertise in the art and science of communication:

*So that's very skilled, and that's very... you only have a short space of time and parents are so traumatised, they are so traumatised at that time... so there's a lot of conversations that are needed in a short space of time. [P1].*

The healthcare professionals used their expertise to overcome such challenges so that they could support children and families.

Providing such support and expertise was not without significant challenges. The healthcare professionals spoke of the importance of devoting significant time in addressing the needs of the child and in supporting parents and families. The healthcare professionals also spoke about the need to intensify this care at end-of-life, this they termed 'stepping up'. Hence, 'Addressing the needs of the child', 'Supporting parents and families', and 'Stepping up' became the subthemes that made up the overarching theme of 'Being there'. The theme 'Being there' and its subthemes are diagrammatically represented in Figure 10. The sense of dedication to the needs of the child was apparent from each healthcare professional interview. Thus, the subtheme 'Addressing the needs of the child' will be discussed first.



**Figure 10: Diagrammatic representation of Theme 1 Being there**

### 7.2.1 Addressing the needs of the child

This subtheme relates to the prominence placed on 'Addressing the needs of the child' as a constituent of 'Being there'. The common goal of all healthcare professionals was to provide optimal care and comfort to the child throughout the care trajectory regardless of duration:

*We focus on what this care means ....we focus on life... every life is valuable, and it's not about the length of time that they're here, it's more about the fact that they are here, and about supporting them. [P1].*

Through their many interactions with families the healthcare professionals regularly developed an intimate knowledge of the child and their care needs. For many healthcare professionals, knowing the child and the nature of the condition was viewed as imperative in being there:

*So, if the parents ring me today and tell me that the seizures are this way or that way or the frequency has increased, I will know. I suppose I have such close contact with the families. I know their child's medications. I know their history. Depending on the nature of this child, I can get them seen within a day or two or we can make a change that day. [P7].*

Providing comfort from symptoms appeared to span across the entire care trajectory. Knowing the child meant that the healthcare professionals could respond immediately and appropriately to their care needs:

*You have to know them. Like one particular child recently, he was just very quiet...just quiet. It wasn't like him. He's arms and legs would usually be constantly going. He was just really quiet, and then he went just that little bit greyish. He had no temperature and from a respiratory point of view appeared well. I knew and the other nurses knew he wasn't right. He ended up with bilateral pneumonia and in hospital for nearly a week. [P12].*

The healthcare professionals felt more comfortable and confident in providing care when they knew the child.

In addressing the needs of the child, all healthcare professionals incorporated into caregiving the belief that the child's parents and family are the constant in the child's life: *"if we think about all these families, they still do a huge amount of the caring themselves."*[P11]. By including the parents and family in the care process they believed that the child received higher quality care. Additionally, due to the nature of the conditions the majority of the children were reliant on their parents' knowledge and observational skills to interpret their care needs. The healthcare professionals fully supported parents in the care process and acknowledged the value of their expertise. The child's voice was also central in the whole process of care. Observing the child and allowing the child the time to communicate their needs was viewed as essential:

*I've seen babies with very little brain tissue, and they can communicate their needs. They can communicate pain, distress, discomfort. So, part of it is just letting the baby dictate the way ... and I find that very much with all the children I work with. [P1].*

By including the child's voice in the care process it also allowed the child to become part of the decision-making process and to dictate their unique trajectory:

*They'll decide their journey. They know their own trajectory, and how their condition will go.*

*They decide. They do decide, and sometimes it's listening to somebody who can't speak. [P1].*

The healthcare professionals truly acknowledged the value of the partnership between themselves and the child and family.

In addressing the needs of the child providing care at end-of-life was often challenging. The child's care needs regularly became more profound at end-of-life. Symptom management in particular was at times very skilled and very complex:

*In many cases their symptom management at end-of-life is much more complex even than... I mean, I'm not minimising the complexity of symptom management for children with cancer, not for a minute, but the complexities for the children that are poorly, improve, poorly, improve, yes, need maybe more technology. That has changed a lot. [P3].*

Furthermore, children with NMLLCs often remained in the terminal phase for a considerable length of time. Managing symptoms was a priority for the healthcare professionals in order to provide and maintain comfort at end-of-life. Anxieties surrounding symptom management were common amongst healthcare professionals who were not regularly engaged in the provision of CPC:

*It's often very neonatal doses. Even to get them to feel confident giving Paracetamol or Oramorph. And that it's okay to give medication to a baby that is obviously distressed, in that its heart rate is increased, it's very cyanotic, so even supporting the professionals there's that. [P1].*

As a result, these healthcare professionals welcomed input from healthcare professionals who were experienced in CPC: *"they're happy to hand over that care, when they know that they're out of their depth."* [P1]. Education and support was seen as essential for healthcare professionals who are not regularly engaged in the provision of CPC so that they feel confident and proficient in addressing the needs of the child.

In addressing the needs of the child, the healthcare professionals identified the systemic impact of NMLLCs on parents and family members. Caring for a child for a long period was considered particularly difficult for parents and family members:

*They have limited time, limited energy, so there's a strain on their relationship. There's limited time for siblings. Of course then it's their grief, their anticipation of what is going to happen to their child. In the case where the trajectory is unpredictable, they have to grapple with this idea that they will have huge care demands potentially over a long period of time. At the same time they're terrified that they're going to lose their child so it's this rock and a hard place.*  
[P10].

It was also perceived as remarkably difficult for families of children whose life expectancy is short. Thus, 'Supporting parents and families' was similarly regarded as a vital component of 'Being there'.

### **7.2.2 Supporting parents and families**

This subtheme relates to the emphasis placed on supporting the emotional and practical needs of parents and families throughout the care trajectory from diagnosis through to end-of-life care. The healthcare professionals had incredible insight and compassion for families and recognised the practical and emotional chaos in which they lived. As a result of having such insight and compassion they were highly responsive to the families' needs at each stage in the trajectory. Communication appeared to be the vehicle through which the healthcare professionals could approach and reach the parents and families in need of care.

The healthcare professionals' narratives reflected skilled communication around diagnosis/prognosis, possibilities of withdrawing ventilation and about possible death tailored to fit the unique needs and biographies of each family and their situation. The healthcare professionals utilised a constellation of communication practices. 'Being there' was of utmost importance regardless of the communication practice employed. Initial communication following diagnosis or recognition of a life-limiting condition was centred on assisting parents and families gain an understanding of the situation and in some instances deal with the practicalities of the situation:

*Well, it's obviously a shock, as you can imagine. You are there with parents and they have this beautiful baby and they are given a dreadful diagnosis. At that point they need somebody to sit with them. I find that being there at that time is very important.* [P9].

The healthcare professionals outlined the importance of spending significant time with parents and families at the time of diagnosis or recognition of a life-limiting condition in an effort to support them:

*So I think sometimes, the more time you have to repeat a conversation. Then you can kind of gauge what the parents are like and what they need, because everyone's different. Sometimes you need to shut yourself down as well, to protect yourself, I think, as a parent.* [P9].

The healthcare professionals were mindful that the high levels of distress experienced by parents and families often impacted on their ability to retain information at this time. Hence, the information provided was tailored to meet the unique needs of the parents and families.

Following diagnosis or recognition of a life-limiting condition the support needs of parents and families waxed and waned and was often dependent on the child's health status. Communication was therefore viewed as a dynamic process taking into account the complexity of both the child and family:

*So when we start to see those deteriorations around feeding, around breathing, maybe seizures, I would have conversations with the families around... nobody knows how long this journey will be. And there will be the ups and downs through the whole journey. And sometimes it's getting to that level and talking to parents about life but it's not always as simple as that, because there's some families that ...life is so busy, busy, busy, that it takes a while, and its working at their pace to a point, but also to start sewing the wee seeds. [P1].*

In communicating with parents and families it was important to provide clear information. Clear information included being honest and truthful with families about the likely process of end-of-life for their child while also acknowledging prognostic uncertainty. Although it was important to prepare families for the future, it was also important to focus on the present and the positive aspects of the child's life: *"sometimes parents will latch onto the fact that they're really, really sick. And they are. Sometimes they need support also to put it aside a little bit."* [P12]. Focusing on the present was viewed as essential in supporting parents and families to live with some sense of normalcy and control.

The changing needs of the child were also important in determining the support needs of parents and families:

*It can be very difficult for parents when children reach four/five years of age. They can no longer carry their child up the stairs, or suddenly they realise that they need a downstairs bedroom. They are losing the respite hours from (name of service provider), and they're looking for more respite supports and often it's not there. And sometimes, the child becomes quite stable, and they have a realisation this is long-term. [P12].*

Not only had families to grapple with their child's condition, they often had to deal with a lack of services for their child. While the healthcare professionals had a key role in assisting families accessing available services emotional support was also viewed as essential during these times.

Communication and information exchange also played an important role in supporting parents and families in complex decision-making. The healthcare professionals spoke of how the reality of the decisions that parents faced throughout the care trajectory appeared to drain them of their emotional resources. Decisions regarding advanced care plans were perceived as particularly difficult:

*Resus treatment agreements... that can be very difficult for parents. It comes up again and again because resus treatment agreements are reviewed may be on a yearly basis, or based on need then as well. And they can change. [P12].*

The timing of discussions was, however reported as challenging due to the inherent uncertainty associated with NMLLCs:

*...when to bring it up, that's always difficult, but I think if you start talking about it early on, you wouldn't talk about an advance care plan, you would have a conversation about the child's medical fragility, if they were to get very unwell, what would happen, what would we do, and discussing the prospect of ICU care, what that would offer, and I think if you can get the parents to a point where they would start to see that maybe ICU care might not be in their child's best interest, because they might not survive it. [P4].*

Early discussions were often perceived as preferable. After the parents and family were fully informed it was often about affording them the opportunity to discuss the decision; the discussion often helped the families find their own answers:

*Families try to strike a balance between doing all that they can for their child, exploring all options, but then not putting the child through more suffering than they need to. I invite them to think about who is it for? In terms of this decision....In whose interest is this being made? Sometimes parents might see that it's for their child and other times they'll see that to a degree they're making it for themselves. There's certainly no place for judgment in that. It's extremely difficult. [P10].*

The goal of decision-making was to reach a consensus which was in the best interests of the child.

Despite having different roles in CPC service provision all healthcare professionals felt connected with parents and families and focused on providing the best care to those families. Certain professional groupings such as nurses and social workers had a more central role in terms of addressing the support needs of parents and families. Other professional groupings such as General Practitioners regularly referred families to appropriate healthcare professionals or service providers when support was required:

*We don't have the time, to be honest, to be proactive. Most GPs, I hope, would be aware of the situation and would know where to direct parents, and you know, deal with it.....depression or stress-related illness for the parents. [P6].*

However, all healthcare professionals spoke about the need for a new more intense approach to care as end-of-life drew near, this they termed 'stepping up'. Thus, 'Stepping up' arose as a subtheme as it was a key component in 'Being there'.

### **7.2.3 Stepping up**

This subtheme represented the belief that there is a need to intensify support for children with NMLLCs and their families at end-of-life. The provision of quality end-of-life care was undoubtedly viewed as paramount by all healthcare professionals. Not only was it important for the child and family during the end-of-life period, it was also important for the family through to bereavement and thereafter:

*Like the last family, their boys were very young. It wasn't just for the boys at that stage, but it was for them right through into adulthood. That was their first experience of bereavement, and it set the tone for the future. [P3].*

The healthcare professionals viewed the outcome of such care as long-lasting and were therefore highly responsive to the needs of the families at this time.

Where possible, end-of-life care was planned and delivered in full consultation and partnership with parents and families. Meeting the expressed needs of the parents and families was viewed as incredibly important. At times the parents' wishes were straightforward and could be easily accommodated:

*The mum had everything prepared, and we talked about if the baby only had a few minutes or no minutes at all. Instead of using the hospital receiving blankets she had bought pink towels and she wanted to use them instead of the receiving blankets. [P3].*

There were times however, when meeting and supporting the parents' wishes were more difficult to accommodate. Often the healthcare professionals were faced with the challenge of arranging complex transitions between hospital and home over a very short duration in order that end-of-life care could be provided at home. Nonetheless the healthcare professionals made every effort to overcome this challenge:

*We would pull out all the stops. Some parents do want to take their baby home. So, we have to pull out all the stops if we know we've only got a very short period of time. So, we would. [P5].*

This regularly meant engaging with numerous service providers and healthcare professionals who all had a unique role to play in the provision of care. The healthcare professionals spoke almost with a sense of relief that their requests for medical cards and essential equipment were responded to in a timely manner and with a minimum level of bureaucracy at this time:

*I've had a medical card granted within an hour. So this would be very much at end-of-life for a child either going home from hospital that has none or for a family who are waiting for a medical card to come through and there's no urgency, but suddenly, the child deteriorates and it is needed very quickly. Yeah, I've never had to beg. [P7].*

The timely response was important in enabling the child to be cared for comfortably at home at end-of-life. The healthcare professionals perceived that service provision in general intensified at this time: *"I find that every service steps up at end-of-life."*[P7]. The healthcare professionals valued the commitment and enthusiasm that existed within the multi-disciplinary team at end-of-life and how they equally 'stepped up' to meet the needs of the families.

On the other hand, the healthcare professionals highlighted that services were regularly cut as soon as the child dies. The end of relationships with healthcare providers who had been with a family for a long time was particularly hard for parents:

*Well, isolation is a very common experience. Where you see it in a really marked way is when the child dies. Usually families will have contact with several services that end all at once. That's when you really see that isolation. [P10].*

The healthcare professionals once again 'stepped up' and adapted and responded to the needs of parents and families at this time:

*The family want someone who knows their child and if you're visiting, they want you to go to the grave, they want you to look through pictures, and they want you to go through everything that happened. They often want to talk about regrets. [P7].*

The healthcare professionals were cognisant that this support needed to remain 'stepped up' following the death of the child.

Hence, 'Being there' exemplified expert care that was individualised and responsive to the unique needs of children with NMLLCs and their parents and family throughout their care trajectory. Three components were essential in 'Being there' those being 'Addressing the needs of the child', 'Supporting parents and families' and 'Stepping up'. The provision of expert care was certainly challenging for all healthcare professionals. Additionally, it was often threatened and compromised

by the intricacies and challenges associated with CPC service provision. Consequently 'Being focused' on such care was truly essential.

### 7.3 Being focused

This theme captured the sense that the healthcare professionals remained entirely focused on providing expert care despite the multiple intricacies and challenges involved in CPC and service provision. As previously stated the healthcare professionals had a variety of roles in CPC. In addition, they delivered care in a variety of settings including hospital, community, hospice and respite settings. Regardless of professional role or setting in which care was delivered all healthcare professionals encountered many challenges in the delivery of such care. Despite these challenges the provision of expert care was beheld by all as immensely valuable:

*I think if they are getting that support from us in the hospital when their child is here and that support is extended to the community and extended to the family home. I think we can make their burden just a little easier. [P11].*

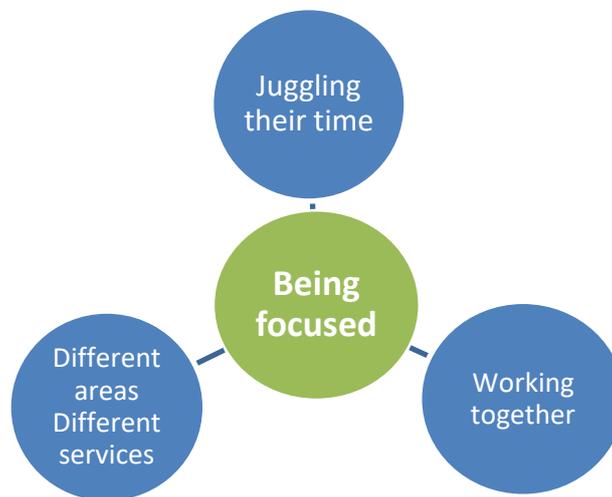
Remaining focused on the care provided took precedence over any challenge encountered.

Although time was emphasised as an important factor in the provision of expert care the majority of healthcare professionals struggled with time constraints. As a result they regularly pushed themselves by juggling their time to be there for all. While the healthcare professionals were touched by how parents managed and had a sense of admiration for them, they also articulated unease about the vulnerability of parents within this role as well as concern with regard to the level of recognition and support they received:

*they do a huge amount of the caring themselves; they get very little time to themselves. Some of them are taking care of the most complex of children in this country, in their own home and very often with limited supports. [P11].*

The healthcare professionals emphasised the importance of providing a coordinated and collaborative CPC service in order to meet the needs of families: *"I definitely think for those parents to survive and keep doing the job that they do every day, they need support. There's no two ways about that."*[P5]. Many healthcare professionals spoke of the efforts they had made in their service area to provide a coordinated and comprehensive service. Yet the frustration of the healthcare professionals regarding CPC service provision was evident throughout the narratives. The healthcare professionals identified major gaps in service provision. Furthermore, different areas had different services, making it difficult to meet the needs of families depending on where they lived.

Additionally, while the importance of working together was reiterated during the course of the interviews this was often tremendously complex. Therefore, 'Juggling their time', 'Different areas Different services' and 'Working together' became the subthemes that made up the overarching theme of 'Being focused'. The theme 'Being focused' and its subthemes are diagrammatically represented in Figure 11.



**Figure 11: Diagrammatic representation of Theme 2 Being focused**

### **7.3.1 Juggling their time**

Against a backdrop of limited time and resources, the healthcare professionals remained entirely focused on providing expert care throughout the care trajectory. The healthcare professionals regularly prioritised their time in order to meet the needs of families who were most in need of care. The following quote resonated sentiments expressed by a number of healthcare professionals: *“it’s not that I’ve lots of time, but you make lots of time.”*[P9]. For some, this meant working beyond their designated hours in an effort to support families. Often services were organised on a 9am to 5pm working day basis, however in reality, this did not meet the needs of the families: *“I’ve never been finished at five ever, because lots of things happen in the evening. It’s often in the evening when parents need the support.”* [P7]. Many healthcare professionals were of the opinion that appropriate support was not accessible for these families if they themselves were not available: *“they need an out-of-hours phone service. Yes, somebody who knows the child.”* [P7]. The healthcare professionals regularly made themselves available to respond to crises due to a lack of available supports:

*Well, I suppose I would leave the phone on at the weekend, and I would check it once or twice, just to see. But families would very rarely ring me at the weekend. The staff on the ward would ring me if they weren't sure about something. [P5].*

The healthcare professionals felt that they could offer simple and convenient access to advice so that appropriate care could be delivered quickly and efficiently. The absence of annual leave cover was repeatedly challenging for healthcare professionals:

*You're trying to prevent that crisis from happening, but you can't prevent every admission from happening. When we go on holidays, we dread it. You are making plans with parents for weeks and weeks up to when you go on holidays as to who they will contact. Again, going through A&E, it can be a very difficult process. [P7].*

The healthcare professionals were mindful that despite careful planning with parents that, unexpected deterioration could occur. They were also mindful of their own limitations and how at times they needed to hand over the care. Time constraints were significant for healthcare professionals throughout the care trajectory. However time constraints were particularly significant following the death of the child and into the bereavement stage.

For many healthcare professionals, time constraints impacted on their ability to provide what they perceived as adequate bereavement follow-up support. This was particularly evident in the hospital and community setting. With the number of children in their care and the wide geographical area that needed to be covered, it was difficult for some healthcare professionals to allocate sufficient time for bereavement follow-up support. Furthermore bereavement follow-up support was both demanding and time-consuming:

*When you're following up with a family whose child has died, it's not a two hour visit. It's an actual day. And I've learnt from my experience where I've had other families booked in to visit in the afternoon or evening, there's no point. [P7].*

Many healthcare professionals spoke about how they had initially struggled with estimating how long a bereavement follow-up visit could take. Furthermore, the duration often varied depending on the families' readiness to engage and their emotional state. Regardless, the healthcare professionals focused on doing the best that they could within the time constraints they had to support families during the bereavement stage:

*I would certainly get one or two visits in and then it would be phone calls, but I find it very hard following up because it takes time. Sometimes with the new referrals and with the children who are alive, and the families who you have got to deal with it on a day-to-day basis. Yes, I find that upsetting and I find that upsetting for them. [P7].*

However the healthcare professionals reported feelings of frustration and guilt in relation to not being able to provide sufficient bereavement follow-up support to families:

*They know they can ring you but you know what, they are so kind that they don't ring because they're afraid of disturbing you, because they've been through it, they're thinking you're going through that with another family and they wouldn't take that time from another family. [P7].*

It was apparent that tensions existed between the strongly felt desire of healthcare professionals to provide bereavement follow-up support and their capacity to do so. Following the death of a child they cared for, some healthcare professionals were worried for the parents and knowing that they were coping was comforting. The hospice setting appeared to provide the most optimal work environment with healthcare professionals outlining that they could respond to the individual needs of parents and families and could spend time with families following the death of their child. However, hospice services were not always accessible to families as different geographical areas had different services.

### **7.3.2 Different areas different services**

This subtheme relates to the inequalities that the healthcare professionals' accounts identified in relation to palliative care service provision for children with NMLLCs. Numerous instances of geographical inequalities were described by all healthcare professionals. Across all service areas and even within service areas inequalities were highlighted. These inequalities were viewed as detrimental to care provision:

*The system is very inequitable. If... and I won't even highlight areas...but if you live in one area in this region, you really get nothing as in homecare support and if you live in another area, you'll get quite a bit. You'll get good help but it's never enough, but you'll get help. It's not consistent. It's not standard. No, there's a huge inequitable service. [P7].*

The healthcare professionals were keen to highlight that they as individuals focused on providing an equitable service: *"I would give the same amount of time to the family that lives two hours away as to the family who lives across the road from the hospital. I would be consistent."*[P7]. However the service as a whole was viewed by them as inequitable.

The healthcare professionals highlighted inequalities in relation to the provision of out-of-home respite care to the extent that some families did not receive any whilst others, in similar circumstances in different areas received a lot: *"in fact, there's no out-of-home respite, for children with life-limiting conditions in this region."*[P4]. Out-of-home respite services appeared to be

located in specific areas meaning that families from outside these catchment areas had fewer opportunities to avail of these services:

*Because (name of service provider) do a fabulous job, and it is fabulous for families who live close by; but if you're going to pack up the car with all the equipment and medications that the child needs, and travel four hours, the parents are exhausted and the child is exhausted, and it just really does not... it just doesn't work. [P5].*

Additionally, accessing out-of-home respite care often proved problematic. Certain known established providers did not appear to have agreed acceptance criteria in place:

*I have made referrals to (name of service provider), but they have refused them. So it's very hard. I have spoken to them about this. You are talking to a family about the service. It's like talking to a family about advanced care planning. You're building them up for this, you're getting them to okay you to make the referral and then you get a refusal. [P7].*

In certain areas the healthcare professionals reported feelings of defeat in relation to this to the extent that they often stopped making referrals. They felt that the process was time consuming and unless they were sure that the referral would be accepted, they weren't prepared to broach the discussion with families. The healthcare professionals also highlighted inequalities in relation to the provision of in-home respite care. The provision of in-home respite care appeared to be largely dependent on the voluntary sector:

*Even talking to other nurses round the country, and some places have a lot of voluntary bodies who provide services. We don't. But in this day and age, we shouldn't be depending on voluntary agencies. Really, the government need to look at the whole issue. [P5].*

Once again different areas had different services available for families. Additionally, accessing healthcare professionals to care for children who required in-home respite care was more challenging in certain areas (such as rural regions) than others.

Inequalities were also highlighted in relation to therapeutic interventions for children with NMLLCs:

*Some children get physiotherapy and occupational therapy every week, some children once a month, some children every six months. And I've worked in a lot of different counties in Ireland and there are no standards. But some of our children are very well served with their therapy service, but its hit and miss with where you live and who you're linked in with. [P8].*

The absence of national standards in relation to therapeutic interventions for children with NMLLCs was seen as challenging:

*There are no standards, nationally. You know, for physio, OT, speech and language, there are no standards. And if there's an occupational therapist on maternity leave, they're not*

*replaced. So you could have a child with no therapy, say physiotherapy, for a year, because it's not available in that area. So that's been happening the last few years. It's been difficult. The recession has wreaked havoc in many, many ways. [P8].*

The healthcare professionals suggested that not only were there insufficient funding to operate therapeutic services in certain areas, but there were insufficient healthcare professionals to deliver such services. Therapists were not replaced when on maternity leave and annual leave and so no service was available to the child during these occasions.

Psychological services for parents were considered to be especially lacking across different areas. This was despite the fact that all healthcare professionals considered psychological support to be an essential component of care for parents and families. Where services were available, they appeared to be available for specific cohorts of children or located in specific areas:

*It's only if they're long-term hospitalised that they would get the support. I'm not saying they all need counsellors or anything like that, but the psychological care needs are terribly neglected. They really are. There's a definite need there for psychological support for families. [P8].*

The healthcare professionals viewed parent support groups as an important source of support for parents. They considered that membership of a support group could buffer the stress, reduce isolation and assist parents to navigate the daily challenges inherent with having a child with a NMLLC:

*I think parents want to link with other parents and families. Sometimes they're very isolated, like no one will understand. They want some empathy. They're very effective in that way. The information exchange between parents is also very effective. [P10].*

However support groups were often geographically scattered, therefore access was not always available for families.

In most service areas the demand for and supply of certain equipment presented challenges to healthcare professionals, leading to requests having to be justified and then approved by managers, increasing the length of time to get equipment into the community:

*Equipment, it's a nightmare. Children are often waiting for buggies, waiting for standers, waiting for bath seats. I've been in a house where this beautiful high-low chair arrived, finally, and the child wouldn't fit into it because she was too big. [P8].*

The high administrative workload increased pressure on the healthcare professionals' time. Regularly experienced healthcare professionals spoke about how they found it difficult to navigate the system in relation to equipment. However, in some areas this did not pose such a challenge:

*I know everybody down to the suction guy, down to the oxygen guy, I've met them all. [Laughter] I've introduced myself. We all know each other. I have met the area disability managers. I have met the aids and appliances crew. I meet the person who signs off for the... I have met everybody. And I will be very clear that I will not look for something unless it's really necessary. [P7].*

In certain areas the healthcare professionals had fostered excellent relationships with administrators and suppliers. Due to the range of healthcare professionals and agencies involved in CPC, the importance of working together was reiterated during the course of the interviews. In reality, working together was often extremely complex.

### **7.3.3 Working together**

This subtheme relates to the importance placed on 'Working together' in order to deliver expert care. With the number of healthcare professionals and service providers involved in CPC 'Working together' was however complex and multi-faceted. Nonetheless, it was viewed as an essential component in the provision of expert care. Effective communication and collaboration were intrinsic in working together. Where communication and collaboration were effective the needs of the child and family remained central however, where communication and collaboration broke down the focus on the needs of the child and the family became somewhat skewed.

In the majority of areas a key worker was employed as a support and liaison person to the family in relation to all aspects of the child's care, which undoubtedly helped in the process of communication and collaboration. All healthcare professionals viewed the appointment of a key worker as positive:

*She's just made it so much easier...the fact that she's able to bridge between hospital and community, and get back to families about queries very quickly. And also, she's done a huge amount of work at engaging all the services with a child. [P4].*

It appeared that the key worker provided the vital link between hospital and community services and was therefore viewed as fundamental in the care process:

*It also ensures that I know what's really happening with the children, because she's in contact daily with the children who are very fragile, and gives me constant updates. And of course, she's very good clinically, and will prompt me to see children if I need to see them, and that's great, so that means we're able to provide a flexible service, and she allows that. [P4].*

However, issues surrounding communication and collaboration were also evident.

The degree to which information and documentation was shared varied among areas, owing to differing IT systems, record keeping protocols and accessibility related to geographical location and organisational supportive infrastructures. There was little evidence of electronic record keeping:

*That was something that struck me when I started here, that it seemed a pity that people weren't able to share, even between institutions and organisations, and pool the knowledge and update it in real-time. That people weren't able to have a cloud that all services could tap-in to and update. That would seem to be a very sensible resource. [P10].*

The absence of electronic record keeping made the process of communication extremely difficult. It appeared that the healthcare professionals utilised every avenue of communication, including face to face, phone, email, whiteboards and handwritten communication in an effort to enhance communication and thus enhance care. All avenues of communication presented their own unique challenges.

Parent-held folders were introduced in many areas. These folders contained up to date and pertinent information regarding the child. The folders were introduced in response to issues raised by both parents and healthcare professionals. The introduction of parent-held folders was generally viewed as positive. The healthcare professionals outlined a number of practical and safety benefits in using these folders:

*Having the medicine list in milligrams and mls for the likes of A&E or for the GP because nobody knows the concentration of medications that they have in the house, and parents often concentrate on mls rather than milligrams. [P7].*

However, they also noted that some parents were reluctant to use the folder particularly if their child was in receipt of services for some time. Communication was particularly challenging in the Emergency Department. The healthcare professionals outlined that the child's chart was traditionally used in the Emergency department. Many key workers included an updated summary in the front of the child's chart which again contained up to date information on the child's condition and treatment regimes. Although this once again added to the workload, it was seen as worthwhile in order to ease the burden for parents who were attending the Emergency Department:

*- So that they don't have to tell their story all over again. Because you have a ten year old in A&E, there are three volumes of charts and the doctors are asking about the child's birth. I mean it just would drive the parents... it drives them crazy. [P7].*

Face to face communication was seen as the only viable method of communication for children who were at high risk and who needed to be assessed immediately if they attended the Emergency Department. The health of many children was so tenuous that minor delays in assessment and treatment could lead to major consequences. Face to face communication often meant that the healthcare professionals had to attend the Emergency Department in the evening time in order to provide the necessary information to night duty nurses. In some areas children with NMLLCs were given direct access to their local paediatric unit which meant that this was not an issue.

All healthcare professionals identified the need for improved communication in order to enhance care for children with NMLLCs and their families. There were many examples of poor information exchange and breakdowns in communication between services. The healthcare professionals reported that poor communication often significantly added to the parents' distress:

*You can never change the way bad news is broken. The impact that that can have on parents can be devastating, it can be life-long as well, and you are trying to get them to recover from that for a very, very, very long time. [P11].*

Memories related to dissatisfaction with the way the diagnosis or bad news was delivered often remained with parents throughout the trajectory and influenced future relationships with healthcare professionals. Therefore poor communication had several potentially negative consequences, for all involved in the care process. The healthcare professionals reported poor communication and collaboration across the hospital-community interface; community healthcare professionals were not always informed of readmissions, discharges or new treatment regimens:

*It's a nightmare. Any meeting we ever go to, the conclusion is that communication needs to be improved. From a GP point of view, there is a delay from the hospital. Seizure management is the big one. Parents phone up asking for such-and-such, anti-epileptic. We've never prescribed it; we didn't know they were on it. Now, parents know, parents will tell us, that their child was in hospital and they've increased the Lamictal or Keppra, or they've juggled this, but a lot of it is relying on the parents' word. [P6].*

Poor communication and collaboration frequently contributed to an increased workload for healthcare professionals in the community. Additionally, concerns were raised that lack of timely communication could result in medication errors. The healthcare professionals were also concerned that there was an over reliance on parents to pass on pertinent information.

Many healthcare professionals were concerned with the lack of communication and collaboration during transition to adult services:

*Transition is a big word, and there's nobody to hand over to because from a hospital side of it, it's whatever consultant is on call. So there is no team that you hand over to. In the community, yes you hand over to adult services in the community. That is much more straightforward. [P7].*

It appeared that adult services were either absent or did not engage during the transition period:

*Transitioning to adult services.....I find that's really difficult, to be honest. Because paediatricians step back, and then there's really nobody then to fill the gap. We have a holistic view of care, whereas once they turn eighteen, or finish school, they're back to their GP, and then they're going to a disease-specific physician, and that's very difficult. They're not going to take the overall view, and also the whole nursing and disability services change. [P4].*

The healthcare professionals often feared that families would be either abandoned or not cared for in a similar manner by healthcare professionals in adult services. A transition coordinator was employed in the hospice setting to coordinate transition to adult services. This was seen as a facilitating factor in the transition process.

Documentation in the home was often difficult to manage. In some homes there could be up to four service providers involved in the child's care. Often each provider used their own individual documentation. In a bid to reduce duplication, a number of regions developed parent-held care plans. The care plans contained key information relating to the child. All healthcare professionals who cared for the child in the community could record their notes in this record. This proved helpful in the communication process. However the healthcare professionals also worried about governance issues in relation to the parent-held care plans. Additionally, certain providers continued to use their own documentation and had not adopted the use of the parent-held care plans. The need for clarification regarding governance of the parent-held care plans was seen as essential. The healthcare professionals anticipated that this issue would be addressed and in the interim, they focused on 'working together' within the constraints identified in an effort to meet the needs of the child and family.

Hence, 'Being focused' captured the sense that the healthcare professionals remained entirely focused on providing expert care despite the multiple challenges and intricacies involved in CPC and service provision. Three elements in particular threatened and potentially compromised the provision of expert care; 'Juggling their time', 'Different areas different services' and 'Working

together'. However 'Being focused' enabled healthcare professionals to overcome these challenges. Nonetheless, both 'Being there' and 'Being focused' impacted both personally and professionally on the healthcare professionals. 'Being strong' was therefore essential.

## 7.4 Being strong

This theme epitomised the personal and professional impact on healthcare professionals who are working in the area of CPC and the manner in which they sustain themselves in 'being focused' and in their 'being'. All healthcare professionals reported that they felt emotionally touched in different ways in caring for children with NMLLCs and meeting the needs of families. The healthcare professionals shared stories, frequently with heartfelt responses about parents and children who they cared for:

*I am emotional as it brings me right back to that situation. For those wee twins, we did canvasses with both boys on it, handprints and footprints and little fingerprints. Those are precious. The mummy got to feed her baby. Only like 5mls of breastmilk at a time. Sometimes not even that. That was purely for that mummy just to be able to give that basic nurturing. There's that physical need to do that with your baby, and she had twins, so she had one bruiser and one baby that was..... but equally, had needs. [P1].*

Many healthcare professionals shed tears in the course of the interviews as they recalled situations and disclosed the lasting influence of their experiences on them personally and professionally.

The healthcare professionals described how they gained perspective with each palliative care experience:

*And it's only little things; it's not even complicated things. A lot of times it's affirming parents around what they are doing and talking about what other families have found beneficial in the past. But it's so valuable. It makes such a difference. [P3].*

The perspective that healthcare professionals gained through their interactions with families gave them the strength and energy to keep going and keep focused. The death of a child was regarded as an emotional experience by all healthcare professionals, but with experience came a level of comfort:

*She caught a chest infection and became clearly very unwell into the night and died very peacefully in her parents' arms at the age of nine months, so it all worked out ideally the way they wanted it. It worked out the way they wanted it. [P7].*

In time the healthcare professionals developed the strength and resilience to deal with anguish and death. They felt rewarded that they were able to support and provide comfort to children and families.

Caring for children with NMLLCs and meeting the needs of families were described as both demanding and complex particularly when it involved insufficient resources. Yet most healthcare professionals described altruistic intentions and despite the challenges, they were able to derive self-motivation and value their role. The healthcare professionals had the strength and vision to see that they had the ability to make a real difference in the lives of these children and families:

*You're going in with just the skills that you have, to deal with that situation, to support the families, the parents, the professionals, and make sure the care is of a standard that – whatever length this life is, that it's valuable, and the child doesn't need to suffer or be distressed. [P1].*

The healthcare professionals strove to deliver high quality care. They had strengths in developing others and found meaning in their experiences. They reported feeling enriched from their work and felt they gained a new perspective on life in relation to what is most important and meaningful:

*This kind of work is very grounding. It's very grounding around just... I suppose the value of life, of your experiences, of your children. I think sometimes, when you get more grounded, you can see the value of it in your own life. [P1].*

Through their many meetings with parents and children the healthcare professionals appreciated and re-evaluated their lives.

The healthcare professionals felt that they were doing the best that they could to support families. However, they also felt a sense of powerlessness and lack of control over workload and patient care. They too needed support in providing expert care. Hence 'Giving my all' and 'Healthcare professional needs' became the subthemes that made up the overarching theme of 'Being strong'. The theme 'Being strong' and its subthemes are diagrammatically represented in Figure 12.



**Figure 12: Diagrammatic representation of Theme 3 Being strong**

#### **7.4.1 Giving my all**

The healthcare professionals demonstrated an extraordinary level of commitment to families and viewed it as a privilege to work in CPC. They were keen to convey their commitment to ‘make a difference’ in their role. This was something they strove for, believing that only their best was good enough. Nonetheless the healthcare professionals often worried about sustaining the care. They were aware that they had to remain strong and thus it was necessary at times to hand over the care. However the lack of resources made this extremely difficult. Not only had they to juggle their time they had to juggle the limited resources such as nursing hours for these children:

*It’s a bit like the loaves and the fish – we can only give from our organisation what we have got to give, but there are times, if families need more, so either times of deterioration or trauma, or that they need more support from us we try and respond to that. [P1].*

This often meant taking hours from one family to meet the needs of another family.

The continuous unavailability of nurses with the necessary qualifications, skills and experience to care for children with NMLLCs in the home impacted significantly on the healthcare professionals. Even when nurses were available their hours were often limited:

*The majority of them are working second jobs. So when they’re working second jobs, they’re not always available to meet you at a given time. I have, I suppose against my better judgement, met nurses at half-ten at night, going to a family because a family is so nervous about this person starting and you want to make that as smooth as possible. Again, making*

*sure the nurse knows what's happening, what they are to do and this is in particular end-of-life cases. [P7].*

The healthcare professionals were often challenged with the responsibility of introducing new nurses to the child and family and providing education on a 'need to know' basis:

*And like, I could go out and train someone to look after a PEG tube. But I'm not going to be there at 2:00am, watching them while they're physically actually doing it. So, it is definitely a huge issue. [P5].*

Often the healthcare professionals found it difficult to trust other healthcare professionals to care for children and families. The healthcare professionals identified the urgent need for clarification regarding clinical governance. Suggestions on how to improve this situation included a bank of nurses with core skills who could be employed to provide care for this cohort of children.

#### **7.4.2 Healthcare professional needs**

The healthcare professionals required knowledge, competence and experience in order to feel confident in their professional role. Many healthcare professionals recalled how they had initially struggled in their professional role in providing CPC. They spoke about how they developed confidence with experience and exposure to children requiring palliative care. They likewise spoke about the importance of education and how it helped them to develop the strength and resilience to work in the area:

*And then by default, and through fear and trepidation, I did the Paediatric Diploma in Palliative Care, which was the year-long course, basically for my own mental health and to reassure myself. It gave me the confidence, as well as just networking and, you know, knowing where to find answers and, yes, just personally, yes. [P6].*

The narratives clearly revealed the strength that the healthcare professionals had in managing their professional development needs.

The healthcare professionals reported that providing CPC was emotionally demanding. They highlighted the moral distress that they and their colleagues occasionally encountered, particularly in relation to end-of-life decision-making:

*The parents really just wanted to keep going no matter what, and it was very, very sad for them because the staff, afterwards, actually felt that they needed support personally, because they found it very harrowing to go in and care for this baby who they felt they were hurting and not helping. And, really, for the parents, of course, who were in a different place and couldn't see that. [P9].*

The healthcare professionals had the strength to detect useful support networks. The majority of healthcare professionals felt able to share their experiences with other healthcare professionals, thus lightening the emotional load on them. The development of supportive professional relationships that promote debriefing was viewed as important. The healthcare professionals appeared to rely on colleagues more than family or friends for support. Interestingly, some healthcare professionals referred to being lucky when reflecting on available support:

*Luckily, see, the lovely thing about neonatal intensive care is there's usually a group. In the middle of the night... if I have a very difficult decision, I do have colleagues that I would ring....Even if it's late at night. [P9].*

This was interpreted as having professional support is never a guarantee.

As ethical conflicts were often a source of stress for healthcare professionals they identified the need for self-awareness with regard to the provision of CPC:

*We have to look ethically at what we do. We are looking after these children, and we're keeping them alive, and then sometimes you have to look at where the endpoint is. Do you keep on pushing and pushing and pushing? Are you doing that for the parents? Are you doing it for the child? Are you doing it for medical perfection? Because often the consultants and doctors are very much curative in their perspectives, and often it's difficult to get through that palliative care ethos. [P5].*

The healthcare professionals regularly reconsidered their own values in an effort to sustain themselves. Throughout the interviews they offered insight into what they perceived as 'quality of life':

*I'm not talking about not smiling and so on, but uncomfortable and painful quality of life -I'm thinking of the child I was talking about earlier, who never really reached a six week milestone, but looks really comfortable, is growing beautifully, has loads of love, is very well cared for, versus a child who... we have to sedate all the time because they're in so much pain; they can't communicate, have a lot of complications requiring lots of surgeries. [P9]*

Reconsidering their values and considering what they perceived as quality of life was significant in sustaining themselves in 'being focused' and in their 'being'

Hence, 'Being strong' epitomised the personal and professional impact on healthcare professionals who are working in the area of CPC and the manner in which they sustain themselves in 'being focused' and in their 'being'. Two components were essential in 'Being strong' those being 'Giving my all', and 'Healthcare professional needs'. The theme 'Being strong' was clearly interlinked with

the two other themes 'Being there' and 'Being focused'. While 'Being there' explicitly highlighted expert care that was individualised and responsive to the unique needs of children with NMLLCs and their parents and family throughout their care trajectory the provision of such expert care was often threatened and potentially compromised by the intricacies and challenges associated with CPC and service provision. 'Being focused' captured the sense that the healthcare professionals remained entirely focused on providing expert care despite these challenges and intricacies. 'Being strong' without doubt enabled the healthcare professionals to 'be there' and 'be focused'. The three dominant themes and eight subthemes that emerged charted the healthcare professionals' experiences of caring for children with NMLLCs, including their experience of service provision and decision-making surrounding the child's care.

## **7.5 Concluding comments**

This chapter has illustrated the healthcare professionals' experiences of caring for children with NMLLCs, including their experience of service provision and decision-making. While the experiences of healthcare professionals caring for children with NMLLCs were distinctive, in many ways commonalities were also apparent in their experiences. The healthcare professionals became a presence in families' lives in several ways and in different settings, circumstances and crises. The healthcare professionals encountered complicated challenges in their work. Organisational structures played a key role in service delivery. The healthcare professionals demonstrated their commitment to deliver the care required by children and families, but against a background of issues relating to working in under-resourced services. The healthcare professionals described the work as emotionally demanding and multi-faceted. Education and support were viewed as essential for both healthcare professionals who are not regularly engaged in CPC and for experienced healthcare professionals. In the next chapter the key findings to emerge will be critically discussed with reference to the initial study aims and objectives and set in the context of the current evidence base.

## **Chapter 8: Discussion of Findings: Healthcare Professionals**

### **8.0 Introduction**

In this chapter the key findings to emerge will be critically discussed with reference to the initial study aims and objectives and set in the context of the current evidence base. The discussion will be presented under each of the two objectives pertaining to healthcare professionals. Accordingly, the chapter will begin with a discussion surrounding the experiences of healthcare professionals caring for children with NMLLCs including their experience of service provision. The discussion following this will provide an insight into decision-making regarding the care and management of children with NMLLCs. As previously outlined (see Chapter 2) a theoretical base was needed to underpin this study. The amalgamation of the FMSF, Paterson's Shifting Perspectives Model of Chronic Illness and Bronfenbrenner's Ecological Theory were considered appropriate. The theoretical framework on which this study was based will be referred to when interpreting the findings. To commence a brief restatement of the research findings will be presented.

### **8.1 Restatement of the research findings**

Three dominant themes, namely 'Being there', 'Being focused' and 'Being strong' and eight subthemes emerged, charting the healthcare professionals' experiences of caring for children with NMLLCs, including their experience of service provision and decision-making. The theme 'Being there' explicitly highlighted expert care that was individualised and responsive to the unique needs of children with NMLLCs and their parents and family throughout their care trajectory. The provision of such expert care was often threatened and potentially compromised by the intricacies and challenges associated with CPC and service provision. 'Being focused' captured the sense that the healthcare professionals remained entirely focused on providing expert care despite the multiple intricacies and challenges involved in CPC and service provision. 'Being strong' epitomised the personal and professional impact on healthcare professionals who are working in the area of CPC and the manner in which they sustain themselves in 'being focused' and in their 'being'.

### **8.2 The experiences of healthcare professionals caring for children with NMLLCs including their experience of service provision**

Caring for children with NMLLCs and their families was characterised as emotionally intensive and multi-faceted. Relational practice was integral to the experiences of healthcare professionals.

The concept of relational practice broadly refers to the art of authentically being with people and includes reciprocity (Bjornsdottir, 2018; Magen and DeLisser, 2017). Notably the concept of relational practice and family-centred care are closely associated (Foster and Whitehead, 2017). Family-centred care may be understood as working collaboratively with parents and families in caring for ill children (Coyne, Holmström and Söderbäck, 2018; Jakšová, Sikorová and Hladík, 2016). Additionally the concept of liminality was likewise significant to the experiences of healthcare professionals caring for children with NMLLCs. Van Gennep (1960) introduced liminality as a concept within anthropology when writing his seminal work *The Rites of Passage*, which considered transitions in which people undergo within life such as parenthood. Liminality describes those on the threshold of a new social position and rituals that bring meaning to such change (Ganguli-Mitra et al., 2017; Tierney et al., 2013). The research findings will be discussed initially in relation to these pertinent concepts.

### **8.2.1 Relational practice**

Relational practice was integral to the experiences of healthcare professionals caring for children with NMLLCs and was unveiled in the three dominant themes, 'Being there', 'Being focused' and 'Being strong'. As with reflective practice, being relational requires a conscious awareness of both one's own experience in the moment and the experiences of those with whom we work, be they students, patients or clients (Ferguson, 2018). Relational practice is a way of being that includes collaboration, trust, compassion and empowerment (Fyers and Greenwood, 2016). Flyers and Greenwood (2016) suggest that true compassion is based on empathy, respect and recognition of the unique individual and a willingness to engage in a relationship with them that acknowledges the limitations, strengths and emotions of all parties. Thus, it requires that healthcare professionals engage in a 'real' dialogue with clients in this instance, parents and families based on honesty and courage. Understanding clients in their context, what they need and how you can best respond to that need are part of relational practice (Fyers and Greenwood, 2016). Doane and Varcoe (2015) remind us that relational practice is at the heart of practice, which is complex work carried out in rapidly changing situations with diverse individuals and groups of people.

The healthcare professionals experienced that the relationship they built with children, parents and their families was key and it was a major source of both personal and professional fulfilment. They evidently acknowledged the uniqueness of each family and their situation. Regardless of practice setting all healthcare professionals shared stories about the variations in families that they cared for. The healthcare professionals constantly approached the family through a lens of inquiry. This meant

getting to know the children, parents and families and what was important to them in the palliative care process. The healthcare professionals experienced that they could then offer help in the most meaningful way possible. Furthermore, the healthcare professionals' felt that their understanding of the desires and choices of parents and families formed the basis for the strong advocacy role that they assumed. In this role they carefully watched over the quality and continuity of care that the child and the family received. Hence, 'knowing' was experienced as part of a proactive ethos which facilitated care delivery, which was flexible and inclusive. The concept of 'knowing' is linked with relational consciousness and associated with more humanistic care (Branch, 2014). In knowing the family and being known the healthcare professionals opened relational space for families (McLeod et al., 2010). Within this space parents and family members were supported in exploring and understanding the meaning of the illness in their lives and discovering ways of living with, understanding and managing the new demands associated with it. This finding accentuates that the use of the FMSF developed by Knafl and Deatrick (2003) in evaluating can assist healthcare professionals in planning tailored interventions that maximise child and family outcomes. In addition, this finding underscores the importance of the subtle and complex work that healthcare professionals do in constructing relational space for families of children with NMLLCs. The healthcare professionals' relational practice was regularly strengthened in scenarios where relationships existed with families over a longer period. The findings from this research also illustrate the complexities that healthcare professionals encounter in striving to develop a close working relationship with the child and family in what can be a short period of time. Clearly trusting relationships could be formed within a shorter period. In this study, the healthcare professionals' engagement with families included not only the initial joining with a family, but also reflected a commitment to continuing and fostering a relationship to care for the child and the family after the child's death. Evidence of this was predominantly seen in the themes 'Being there' and 'Being focused'.

The healthcare professionals reported that they suspended their own judgement about parents, especially when parents were different from them, for example, in ethnicity, values or beliefs because they recognised the potential of their own assumptions, opinions and judgements in influencing a situation. In so doing, they could concentrate on adjusting to the parent's perspective. The importance of a non-judgemental approach has been previously highlighted by healthcare professionals within CPC literature (Bluebond-Langer et al., 2013; Hoell et al., 2017). In one particular study clinicians described a non-judgemental attitude as paramount in enabling parents of children with life-limiting conditions to feel comfortable enough to express their needs in particular

surrounding end-of-life decisions (Delany et al., 2017). The healthcare professionals in the present study also stressed the importance of knowing the child. The healthcare professionals identified that they viewed the child as a unique individual and accentuated that it was important that the child was respected and nourished in the care they received. Their descriptions revealed how they continually sought ways to value the child's potential in view of their limitations. This was particularly marked in the subtheme 'Addressing the needs of the child'. They also believed that the needs of the child could not be understood separately from the family. It is widely accepted that children are affected by and affect those with whom they have relationships with (Harrison, 2010; Roets, Rowe-Rowe and Nel, 2012; Shields, 2010). The parents were viewed as the child's most trusted and valuable resource, thus strategies to support the parents and families were considered crucial. This insight motivated the healthcare professionals to provide quality care. Knowing the services and support available to children and their family and ensuring these were implemented in a timely manner were also experienced as important.

The significance of the relationship between healthcare professionals and families of children with life-limiting conditions has been previously emphasised (McCloskey and Taggart, 2010; Price, Jordan and Prior, 2013; Reid, 2013). The relationship capacities found in this study are consistent with those described in the literature, which show that relating to children, parents and families is a caring process that involves values, intent, knowledge, commitment and actions. Bergsträsser, Cignacco and Luck (2017) described the experiences and needs of healthcare professionals providing end-of-life care to children in Switzerland. Similar to the findings of the present study the key aspect in end-of-life care provision was identified as the capacity to establish a relationship with the dying child and the family. In the said study, trusting relationships could likewise be formed even in highly acute situations within a shorter period, for instance on a paediatric intensive care unit (PICU). Besides mutual trust and personal engagement, providing comprehensive and honest information contributed to the quality of this particular relationship. A sustainable relationship, based on familiarity and trust, allowed healthcare professionals in the said study to embark on the process of end-of-life care. The healthcare professionals in the present study voiced that they felt more equipped to provide care in a family-centered manner when families felt comfortable to verbalise their needs and desires. Comparable findings have been reported in other studies (Bergsträsser, Cignacco and Luck, 2017; McCloskey and Taggart, 2010; Price, Jordan and Prior, 2013).

### **8.2.2 Family-centred care**

As outlined previously (see Chapter 1) family-centered care is an approach to the planning, delivery, and evaluation of health care that is grounded in mutually beneficial partnerships among healthcare providers, patients, and families (Johnson, 2016). It redefines the relationships between and among consumers and health providers and directs healthcare professionals to challenge the notion of professional-as-expert (Dennis et al., 2017). It appeared that the philosophy and core concepts of family-centred care were embedded within the healthcare professionals practice (Banerjee et al., 2018; Coyne, 2015; Kuo et al., 2012). This was revealed throughout the three dominant themes 'Being there', 'Being focused' and 'Being strong'. The healthcare professionals were respectful of the parents' knowledge concerning their child. They recognised and openly accredited parents' expertise-not just their technical skills, but their expertise in applying these to their child. They outlined that they did not view themselves as 'experts' with specialised knowledge and skills to solve problems, but as companions to 'be there' to support families on their care journey. This was predominantly evident in the themes 'Being there' and 'Being focused'. As alluded to earlier the healthcare professionals described the necessity of finding out what was important to the family and not focusing simply on what they as healthcare professionals perceived as important. Similarly, in the literature, family-centred care was described as not focusing on tasks alone, but including families in consultations and attending to their concerns, fears and hopes (Dudley et al., 2015). The healthcare professionals acknowledged that the relationship with parents and families is shaped by many factors such as the depth and frequency of the involvement the family accepts or the individual personalities of family members and healthcare professionals.

The delivery of optimal family-centered care by healthcare professionals is associated with higher levels of parental satisfaction with services, better parental and child psychosocial wellbeing, and increased professional morale and job satisfaction (Frost et al., 2010; King, King and Rosenbaum, 2004; Law et al., 2003). The healthcare professionals in this study experienced family-centred care as a dimension of quality healthcare and as a holistic approach to care. In addition, their experiences of providing family-centred care were extensive. Klassen, Gulati and Dix (2012) described the work-related demands and rewards in relation to working closely with parents of children with cancer in Canada. The healthcare providers found the establishment of close or long-term relationships with parents throughout the entire cancer journey as rewarding. They also viewed it as challenging to work with complex families. For example, they described challenges that often accompanied working alongside acrimonious separated or divorced parents. They likewise viewed it as difficult to work with parents who were rude or angry or parents who had differing

views regarding treatment and palliative care options. Yet the involvement of parents working in partnership with healthcare providers reflects an approach to healthcare delivery in Canada (McKechnie, 2016; Davies et al., 2017). Evidently these findings were refuted to a certain extent in the current study.

### **8.2.3 Liminality**

It appeared that the concept of liminality was also significant for healthcare professionals in caring for children with NMLLCs and their families. This was revealed in the three dominant themes 'Being there', 'Being focused' and 'Being strong'. Liminality, traditionally associated with transitions and thresholds reflects a position of being in 'limbo' or no-mans-land (Turner, 1967). The concept relates to the 'ritual space' in which one is suspended or wavering between two worlds (Charon and Montello, 2004). It is associated with ambiguity and angst as people experience a shift in their social position. The healthcare professionals acknowledged that the parents and families were regularly struggling to 'find their place'. They alluded to how liminality was manifested in the parents' everyday life and how they tried to organise a self that engaged with the liminal space between caring for their child and their impending death. The healthcare professionals reported that they supported and provided direction to families as they tried to organise themselves in this space. This was clearly evident in the themes 'Being there' and 'Being strong' and specifically within the two subthemes 'Supporting parents and families' and 'Giving my all'.

The application of the anthropological concept of liminality has recently been made in adult palliative care (McKechnie, Jaye and MacLeod, 2011). Additionally, Carter (2017) suggests that palliative care for infants, children and adolescents encompasses numerous transitions and thresholds of uncertainty that are challenging. Healthcare professionals have opportunities to be more comfortable amid such challenges, or perhaps even overcome them, if they are attuned to the unique times and places in which families find themselves throughout the care trajectory. Jordan, Price and Prior (2015) described how parents were in a state of liminality, an unsettling period of transition between one social state or status and another when caring for their dying child. Importantly the findings from the said study highlight the sustained or chronic nature of the parents' liminality. The concept of liminality gives validation to the family being 'stuck in places betwixt and between' a past rich life with relationship and purpose and an acute, chronic or critical illness. Liminality provides a framework for addressing the unbound spaces that families occupy: what is past is behind-the present place is tenuous and temporary and what is ahead uncertain. This place is where the healthcare professionals can offer families guidance (Carter, 2017). Thus an application of

liminality might also be recommended in CPC where matters of uncertainty in a child's prognosis and associated planning and decision-making with parents are addressed (Haward and Lorenz, 2011; Noyes et al., 2013). Indeed, Jordan, Price and Prior (2015) suggest that a structural failure to recognise liminality can impede parents' effective transition. Based on their experience the healthcare professionals in the present study found that conceptualising care in terms of liminality offers a powerful framework for understanding the experiences and challenges faced by families of children with life-limiting conditions as they negotiate this particular life change.

The aforementioned concepts of relational practice, family-centred care and liminality were clearly apparent within the healthcare professionals' stories and appeared to give rise to the very clinically competent expert within this specialised field of practice. Whilst the healthcare professionals appeared to view the parents as expert in their child's care, they also recognised their own knowledge and expertise in overall care provision. Ghanem et al. (2018) suggest that experts differ from novices with respect to both their knowledge base and the epistemic activities in which they engage. The healthcare professionals use of expert knowledge to provide anticipatory guidance, approach problems with creative solutions and recognise that planning is needed before a crisis occurs was evident across the three dominant themes. In addition, Paans et al. (2013) propose that experts communicate effectively in complex situations. The healthcare professionals described experiences in which they communicated with parents and families about the details associated with escalation of care, for example transfer to the Intensive Care Unit. They described how this allowed time for parents to pause and process that information during a chaotic time when the infant/child was unstable. The next section will examine the concept of the healthcare professional as expert.

### **8.3 The healthcare professional as expert**

Experts develop through years of experience and by progressing from novice, advance beginner, proficient, competent, and finally expert (Benner, 1982). These stages are contingent on progressive problem solving, which means healthcare professionals must engage in increasingly complex problems, strategically aligned with their stage of development. Thus, several characteristics differentiate experts from novices. Experts know more, their knowledge is better organised and integrated, they have better strategies for accessing knowledge and using it, and they are self-regulated and have different motivations (Persky and Robinson, 2017). Experts continually focus their effort toward improving their knowledge base and skillset to achieve and maintain their expertise. They must constantly practice, be open to challenging new experiences and engage in self-reflective processes (Persky and Robinson, 2017).

The healthcare professionals in this study were experienced and reported that they felt confident and competent in almost all aspects of their professional role. The necessity of foundational and specialised knowledge emerged throughout the three dominant themes, 'Being there', 'Being focused' and 'Being strong'. The settings in which this knowledge base was acquired varied with each healthcare professional's experience; however, it was clear from the interviews that education needed to be comprised of both theoretical and experiential knowledge for it to be meaningful and relevant. Within the findings, it became apparent that the healthcare professionals did not always feel confident in the knowledge base they possessed prior to providing palliative care to children. This was chiefly evident in the subtheme 'Healthcare professional needs'. A number of healthcare professionals described feeling overwhelmed and underprepared with their initial palliative care experiences. This finding was also mirrored in the literature. Lack of knowledge and education were identified by physicians' as barriers in initiating advanced care planning for children with life-limiting conditions in Australia at the commencement of providing care to this population (Basu and Swil, 2018). The majority of physicians in the said study regarded exposure to advanced care planning and education during training as inadequate. Lyckholm and Kreutzer (2010) likewise identified lack of formal and informal preparation in pediatric palliative care as a major problem. Thus a key finding is that healthcare professionals did not always feel prepared/appropriately skilled to commence their work in this area, but that their cumulative learning gave them experience and helped them gain confidence.

The findings of a study undertaken by Feudtner et al. (2007) revealed that higher numbers of years of nursing practice and more hours of palliative care education were individually associated with higher levels of comfort in providing palliative care and a higher total palliative care competency score. The most significant finding was the association between hours of palliative care education and personal comfort in providing palliative care. This and the aforementioned study suggest that education is a significant factor that can effectively boost healthcare professionals' confidence regarding CPC and therefore should be regularly provided to healthcare professionals. Healthcare professionals working in a children's hospice also identified training needs to help them feel more comfortable and confident in care provision (Taylor and Aldridge, 2017). The healthcare professionals in the present study accepted that CPC is a highly specialised area of healthcare and acknowledged the complexity of caring for children with NMLLCs and their families. For instance, in the subtheme 'Addressing the needs of the child' they suggested that the recognition and management of symptoms in children throughout the care trajectory and as they approach end-of-life can be difficult for even experienced healthcare professionals. This finding was likewise revealed

in previous studies (McConnell and Porter, 2017; Price, Jordan and Lindsay, 2013). The healthcare professionals also acknowledged the changing nature of CPC. Owing to advances in supportive care, some children are surviving often unexpectedly into young adulthood with complex care needs (Kerr et al., 2017; Nageswaran, Hurst and Radulovic, 2018).

The healthcare professionals experienced that the care of children with NMLLCs and their families throughout the care trajectory and in particular, at end-of-life often appeared to result in high levels of anxiety amongst healthcare professionals such as General Practitioners, nurses and midwives who are not regularly engaged in the delivery of CPC. The healthcare professionals in this study reported that they were mindful of this and perceived it as a source of stress. They were primarily conscious that this anxiety may reduce the effectiveness of care given to the child and their family at the very time when it is most important to get it right. Based on their experience they indicated that this created a certain burden on them as they endeavoured to support these healthcare professionals in care provision. This was chiefly evident in the subthemes 'Juggling their time' and 'Giving my all'. The healthcare professionals outlined that they did not expect these healthcare professionals to have specialist skills. They also expressed concerns regarding the emotional wellbeing of their colleagues in these situations. Feelings of inadequacy, emotional distancing and burnout have been reported as being more prevalent among healthcare professionals who are insufficiently supported to carry out their role (Hall et al., 2015; Neilson et al., 2013). Additionally the findings from previous studies revealed that nurses with less experience in relation to providing end-of-life care to children struggled with this role on both an emotional and professional level (Quinn and Bailey, 2011; Reid, 2013). However, because of the relatively small numbers of children with NMLLCs many healthcare professionals engage infrequently in the care of these children. The healthcare professionals reported that this creates challenges in developing and maintaining their skills and knowledge. Again, while the focus of this research is on children with NMLLCs comparable findings have been reported amongst healthcare professionals caring for children with cancer (Neilson et al., 2010). In their study, they also highlight the difficulty in developing and maintaining palliative care skills and knowledge in community nurses due to their infrequent exposure in caring for children with cancer. The subsequent section will examine CPC as a dialectic experience.

#### **8.4 A dialectical experience**

CPC was depicted in this study as a dialectical experience. The healthcare professionals revealed relational practice and family-centred care patterns such as making time and connecting with children, parents and families. 'Being there' and spending time with the child and family gave

healthcare professionals a great opportunity to create trust and provide comfort care. This was manifest in the subthemes 'Addressing the needs of the child' and 'Supporting parents and families'. The healthcare professionals regularly prioritised their time in order to address the social, emotional and spiritual aspects of care. Despite this the majority of healthcare professionals struggled with significant time constraints. This was largely obvious in the subtheme 'Juggling their time'. Many of the healthcare professionals recounted the unpredictable nature of their working day, the juggling of competing demands and the constant need to reprioritise their caseload. The findings of a number of studies likewise revealed demanding workloads with implications such as stress and fatigue for healthcare professionals caring for children with life-limiting conditions (Lewis and Pontin, 2008; McCloskey and Taggart, 2010; Quinn and Bailey, 2011). However the healthcare professionals in the present study seemed to 'find' time, knowing perhaps the way to 'create' time was by being fully engaged with each family in the moments that were available.

The healthcare professionals strove to allocate sufficient time for families and to be flexible with their care needs. They regularly put aside their own personal needs to meet the needs of children and families. This was again evident in the subthemes 'Juggling their time' and 'Giving my all'. The healthcare professionals spoke about being proactive. They endeavoured to address concerns early to limit adverse issues for children, parents and their families. It was also a way for healthcare professionals to maintain their relationship with families. Pontin and Lewis (2008) explored how community children's nurses deliver services to children with life-limiting, life-threatening and chronic conditions in the United Kingdom. The findings suggest that being proactive is central to understanding how community children's nurses work to maintain contact with families. The community children's nurses also spoke about how they endeavoured to respond to issues early to reduce adverse consequences for children, parents and their families. The findings explicate how healthcare professionals give of themselves in their work and go 'above and beyond' the call of duty. A number of healthcare professionals also talked about the necessity of withdrawing particularly with regard to bereavement support. The healthcare professionals attempted to negotiate the tensions between these opposing approaches to palliative care. The healthcare professionals shared descriptions of how their practice setting played a role in CPC service provision. This was mainly evident in the subtheme 'Juggling their time'. The necessity of viewing healthcare professionals' experiences in conjunction with practice and social influences is obvious. The manner in which healthcare professionals interpret the meaning of their work and make sense of the experiences they engage in cannot be done without considering how their workplaces approach the care of children and families and how society views death. Bronfenbrenner's theory reminds us how

important it is to think about the 'big picture' when caring for children with NMLLCs and their families. Characterising complexity in terms of Bronfenbrenner's microsystem, mesosystem, exosystem, macrosystem and chronosystem levels is supported by existing literature in multimorbidity, chronic conditions and general healthcare which demonstrate that complexity goes beyond the individual, includes contextual drivers, and derives from multiple sources for example, system-level and service-level aspects (Shippee et al., 2012; Zullig et al., 2016). Bronfenbrenner's Ecological Systems Theory has also been adapted to provide a novel framework for understanding complexity in specialist adult palliative care (Pask et al., 2018).

Price, Jordan and Prior (2013) investigated health and social care professionals' perspectives on developing services for children with life-limiting conditions at the end-of-life stage in Ireland using issues identified by bereaved parents as priorities. Participants ranked structured bereavement support for families as the most important priority for service development. This ranking was premised on a number of considerations. Firstly, awareness of families' manifest need for bereavement support and lack of relevant services. Second, the consequent pressures experienced, particularly by nurse participants to provide such support in the absence of any dedicated role or training. Significant personal and professional dilemmas arose when families expected bereavement support to be provided, often over the long term, by those previously involved in care. These dilemmas included: feelings of guilt at not being able to provide the support required, burnout and consequent diminution in the ability to provide effective nursing care. Suggestions for alternative models of support focused typically on the creation of dedicated bereavement support workers. While the healthcare professionals in this study reported similar sentiments with regard to bereavement support there were also remarkable differences in their accounts.

The healthcare professionals in this study reported that they felt confident and competent in this aspect of their role. Additionally, while time constraints were a significant concern for a number of healthcare professionals they did not necessarily wish to surrender this role, particularly in situations where they were involved in a child's care over a long period. This was chiefly evident in the subtheme 'Stepping up'. The findings from a study undertaken by Clarke and Quin (2007) may illuminate the rationale for this. For families where the illness trajectory was short, relationships with healthcare professionals were less intense than in circumstances where the child died after a long protracted illness. In the latter case, professional carers articulated the view that families suffered loss at two levels: the loss of their child and the loss of the caring regimen and relationship that grew up around their child. In these circumstances professional carers believed that parents

wanted or needed to return to people who knew their child and who understood the circumstances of their loss. Negotiating these endings was important also for professional carers. Furthermore MacConnell et al. (2013) described the experiences of nurses who provide bereavement follow-up with families after the death of a child or a pregnancy loss. Nurses in this study were also committed to providing ongoing care to families who had experienced the death of a child or a pregnancy loss. Relationships were important to bereavement follow-up care, and the connections with families were often emotional for the nurses.

The healthcare professionals in the present study described the work of caring for children with NMLLCs and responding to the needs of families as a struggle in regard to service provision. This was again evident across the three dominant themes 'Being there', 'Being focused' and 'Being strong'. It involved a juggling act as they sought to provide care in the absence of resources and services or where services were inadequate. This was striking in the subtheme 'Different areas Different services'. Access to services has been previously reported as the most obstructive barrier to quality paediatric palliative care (Bradford et al., 2012). The interviews revealed grave concerns about the stability of the workforce and concerns about providing quality palliative care in light of a distressed and overburdened workforce. The healthcare professionals accentuated that children need to be with their families but they also need skilled medical and nursing support that can be difficult to provide at home. Resources for providing 24 hour end-of-life care, with the ability to rest and relinquish practical and emotional responsibilities were limited, as very small local teams and caseloads were spread over wide geographical areas. In general, it was reported that the system is more reactive than proactive. The healthcare professionals made several suggestions regarding what they perceived as important for parents and families.

## **8.5 Suggestions on what healthcare professionals perceive as important for parents**

Based on their experience of caring for children with NMLLCs the healthcare professionals had suggestions regarding what they perceive as important for parents and families. In addition to adequate services such as respite care, specialist therapies, end-of-life care and bereavement support they clearly perceived that the ability to communicate openly and with compassion were essential to the families' emotional wellbeing. This endorses previous research findings that report the efficacy of effective communication between parents and families and healthcare professionals, especially when serious news must be discussed (Hsiao et al., 2007; Meert et al., 2008). However, these studies have focused on the parents' perspectives on communication. The healthcare

professionals in the current study highlighted strategies that they used to enhance communication. They identified the significance of sharing information with families. The healthcare professionals suggested that they must work with families to process difficult information and provide clarification as needed. They regularly described how they carefully managed the volume, pace and timing of information based on the needs of the parents and families. This was most apparent in the theme 'Being there'. They experienced that information should be shared with mindfulness and without interruption.

The consequences of improper delivery of difficult news were also highlighted by the healthcare professionals. They reported that an insensitive approach increases distress and can exert a lasting impact on parents' ability to adapt and adjust to the situation and can lead to anger and upset. Additionally, they observed that insensitive interactions result in the loss of a families trust and threaten the parents' sense of security, comfort and capability to care for their child. This was marked in the subtheme 'Working together'. Yet the manner in which healthcare professionals choose to deliver difficult news is modifiable through training and behavioural learning (Kiluk, Dessureault and Quinn, 2012; Levetown, 2008). Peng et al. (2017) examined the effectiveness of pediatric palliative care training for pediatric clinicians. After training, participants reported significantly increased confidence in a variety of areas including providing emotional support to children and families, personal knowledge, skills and communication, ethical and legal concerns. It appeared that the effectiveness of the process of communicating difficult news depends on the sensitivity and flexibility to adapt a professional technique to each circumstance, depending on the cultural, social, educational and family contexts. Communication plays an integral role in service quality in all service professions including healthcare professions (Newell and Jordan, 2015). Communication is often taught in a behavioural way, as a set of skills to be mastered; this can be useful for novice practitioners (Weller et al., 2012). However, relational practice requires much more of healthcare professionals in terms of knowing how and when to use the skills (Fyers and Greenwood, 2016). The healthcare professionals in this study experienced that the manner in which they approach parents and families in every encounter make an immense difference to their experience. The emotional facet of caring for children with NMLLCs and their families will be discussed next.

## 8.6 Emotional facet of caring

The healthcare professionals acknowledged that the death of a child had a significant impact on both their personal and professional lives. Notably, they reported that their experience of death and dying was largely a positive experience. The healthcare professionals talked about the rewards they derived in providing comfort to children and alleviating the distress that parents and families experienced. This was apparent in the theme 'Being strong'. They reported that end-of-life experiences positively transformed the way they lived their lives, teaching them to live in the present, cultivate a spiritual life and reflect deeply on the meaning and continuity of life. Previous research likewise suggests that healthcare professionals who provide end-of-life care to children across a range of settings such as hospital and community, also report positive experiences, including rewarding relationships with children and their families and making a difference in terms of reducing physical and psychosocial suffering (Klassen et al., 2012; Reid, 2013). However a number of studies describe the negative impact on healthcare professionals who are caring for children with life-limiting conditions and their families throughout the care trajectory. Indeed the findings from the previously mentioned study undertaken by Reid (2013) described some challenges in maintaining professional boundaries, with some nurses portraying themselves as a 'pseudo-member' of the family. Rather than the younger participants or those with least experience, it appeared to be those without a Children's nursing qualification who struggled to reconcile themselves with the concept of child death.

McCloskey and Taggart (2010) explored the experiences of stress in nurses providing children's palliative care in one region of the UK. In total, four focus groups took place with children's hospice nurses, community children's nurses and children's nurse specialists. The findings revealed that the emotional load of providing palliative care impacted greatly on nurses across all three service areas. The nurses reflected on the consequences of developing deep-seated attachments with children, parents and families, which often moved beyond the expected nurse-patient dynamic and resulted in a sense of loss and grief when the child died. Additionally the findings from a previously mentioned study suggest that although health and social care professionals gained considerable fulfilment from their work, the emotional impact was most frequently discussed in negative terms (Price, Jordan and Prior, 2013). This included strong feeling of inadequacy, frustration and sadness arising from the complex and often protracted nature of professional engagement with dying children, their parents and family. The emotional facet of caring in the present study was also apparent in relation to decision-making.

## **8.7 Decision-making regarding the care and management of children with NMLLCs**

The healthcare professionals in this study detailed how they encountered many challenging and ethical decisions in caring for children with life-limiting conditions. Based on their experience they believed that decisions should be made with the comfort of the infant, child or adolescent as a constant focal point. This was apparent in the subtheme 'Addressing the needs of the child'. The healthcare professionals described scenarios where they advocated for children and families and facilitated parental decision-making, suggesting that they were actively promoting optimal CPC in this regard. The healthcare professionals proposed that the timing of introduction of discussions may impact on choice. They were extremely mindful that events, changes in the child's condition and fears can lead to a shift in perspective. This finding highlights that Paterson's Shifting Perspectives Model of Chronic Illness is fitting to help understand how families approach decision-making. The healthcare professionals also reported the challenges of trying to reconcile parental wishes with conflicting professional and personal beliefs and values. This was marked in the subtheme 'Supporting parents and families'. In some studies, emotional distress in healthcare professionals is described along with the stressors evoking it, for example, having to give care which is not in the best interests of the child most notably if the treatment is expected to cause discomfort or pain (Brandon et al., 2014; Ferrell, 2006; Lee and Dupree, 2008; Pearson, 2013). The healthcare professionals clearly recognised that scenarios of this nature were inevitable by virtue of the particularly vulnerable and fragile patient population they served. They perceived that some of the most challenging and morally driven decisions in CPC involve end-of-life decisions, particularly decisions regarding the use of aggressive, potentially life-prolonging but burdensome therapies. The healthcare professionals employed a variety of coping strategies and identified useful support networks to help them deal with ethical issues. These included self-care activities, engaging in self-reflection and developing a personal philosophy of care. This was manifest in the subtheme 'Healthcare professional needs'. The healthcare professionals frequently reflected on the quality of life of children that survived and the impact on the family.

The findings from this research suggest that the healthcare professionals shared similar ethical perceptions that affirm CPC. However the advancement of technology in itself was occasionally a source of ambivalence among healthcare professionals particularly nurses. Its availability was regarded as a driving force behind the delivery of disproportionate care that leads to moral distress. Despite efforts in collaboration, doctors are still occasionally depicted as upholding the goals of 'saving lives' and 'furthering the advancement of science' often supporting continued technological

interventions beyond the comfort level of the nurse. This was marked in the subtheme 'Healthcare professional needs'. Epstein's (2010) research indicates that physicians and nurses share common goals in neonatal end-of-life care. An overarching obligation of creating the best possible experience for infants and parents was identified within which two categories of obligations (decision-making and end-of-life care itself) emerged. Obligations in decision-making included talking to parents and timing withdrawal. End-of-life obligations included providing options, preparing parents and being with, advocating, creating peace and normalcy, and providing comfort. Nurses and physicians perceived obligations in both categories, although nurse obligations centred on the end-of-life care while physician obligations focused on decision-making. The findings demonstrate that although the ultimate goal is shared by both disciplines, the paths to achieving that goal are often different.

The healthcare professionals described how the culture of the work environment was also significant in dealing with moral and ethical concerns particularly in relation to decision-making. A number of healthcare professionals emphasised the fellowship with colleagues as vital in managing and dealing with difficult situations. This was evident in the subtheme 'Healthcare professional needs'. Thorne et al. (2018) investigated how direct care neonatal practitioners understand and experience moral distress. The practitioners reported that they would have valued opportunities for conversation about the clinical episodes that triggered their moral distress; many explained that, in their opinion the culture of the NICU did not support it. In working out how to reframe their most distressing experiences and regain a perspective on how to reboot their capacity in the workplace despite difficult circumstances, most of these healthcare professionals craved opportunities to share their stories, make sense of their dilemmas, and seek closure with both families and colleagues. In addition, Cook et al. (2012) revealed that nurses often did not feel comfortable discussing their experiences at home, but rather typically turned to colleagues for support.

Rushton, Kaszniak and Halifax (2013) describe moral distress as a pervasive reality of palliative care practice. They suggest that a full understanding of the process can help to mitigate or to avoid the progression of distress and concurrently to appraise the situation that leads to moral distress. McConnell and Porter (2017) explored the experiences of healthcare professionals who provide end-of-life care within a children's hospice. The findings suggest that the hospice setting provides a model of excellence in supporting staff and mitigating challenging aspects of their role, which includes peer/organisational support and regular ongoing training in key aspects of CPC. Generally the pace of working is more relaxed in a children's hospice and the working environment much more pleasant with lots of light, space and outdoor areas (Hunter, 2017). Hunter (2017) described the

nature of work within a children's hospice and in particular the role of a paediatrician working within this setting. A reflective approach is particularly encouraged with the children's hospice. The concept of self-reflection, first introduced by Schon (1983) in relation to the reflective practitioner, continues to gain popularity and is advocated in professional practice literature (D'Cruz, Gillingham and Melendez, 2007; Menard and Ratnapalan, 2013; Sandeen, Moore and Swanda, 2018).

## **8.8 Concluding comments**

The healthcare professionals experienced that the diagnosis or recognition of a NMLLC creates a complex, systemic, life-changing experience that propels families along uncertain trajectories. The healthcare professionals perceived that they have opportunities to mitigate suffering and to find relationship, space and language that works in synergy with families to participate in a reality that is unthinkable and yet undeniable. Practitioner attunement to the child and family is a critical component of CPC and can enhance delivery of CPC services (Koch and Jones, 2018). The healthcare professionals in this study spoke of a willingness to enter into conversations with parents and families in which the outcome was unknown. They reported that fostering conversations among family members and between the healthcare professional and the family expanded possibilities for new understandings to emerge. Organisational barriers to relational practice certainly emerged as an issue in the interviews. The healthcare professionals coped by being mentally focused and emotionally prepared, maintaining boundaries with compassion and reminding themselves that they cannot fix everything all of the time.

## **Chapter 9: Integrative discussion and conclusion**

### **9.0 Introduction**

Whilst the data has been analysed and critically discussed separately in the previous chapters, an integrated approach to the overall discussion will be adopted in this chapter. This is important as it can lead to a deeper insight than separate analyses (Tariq and Woodman, 2013; Tonkin-Crine et al., 2015). The original contribution the present study makes to the existing literature base will be considered. The chapter will also present the strengths and limitations of the study and implications for practice, future research, education and policy. The chapter will commence with a brief description and discussion with regard to how the qualitative and quantitative research findings from the parents and the qualitative research findings from the healthcare professionals were integrated.

### **9.1 Integrating the qualitative and quantitative research findings**

As previously outlined (See Chapter 4), in order to integrate the findings, the findings were triangulated. In this section, in accordance with the recommendations of Farmer et al. (2006) and O' Cathain et al. (2010), the findings across the entire study will be considered in terms of convergence (where data sets agree or are consistent); complementarity (where data sets offer complementary information on the same issue and therefore enhance knowledge and/or understanding of the phenomenon under investigation); silence (where one data set uncovers an issue whereas it appears silent in the other data set); and discrepancy or dissonance (where the data sets have conflicting findings).

### **9.2 Discussion of integrated findings**

In response to the overarching research questions posed by this mixed methods study, the findings were integrated according to three categories. These categories were; 1) experiences, including the experience of service provision, 2) access, and 3) decision-making regarding the care and management of children with NMLLCs. These categories were directly related to the study objectives.

### **9.2.1 Experiences, including the experience of service provision**

The qualitative and quantitative research findings from the parents and the qualitative research findings from the healthcare professionals were predominantly consistent. Caring for a child with a NMLLC throughout the care trajectory was characterised as a multi-faceted experience for parents and families. Similarly, caring for children with NMLLCs and their families was characterised as a multi-faceted and emotionally intensive experience for healthcare professionals. This echoes the findings of previous studies (Collins et al., 2016; Nicholas et al., 2016; Taylor and Aldridge, 2017). Parents and healthcare professionals equally demonstrated an extraordinary level of commitment to the child, focusing on providing expert care. Relational practice was integral to the experiences of healthcare professionals. As alluded to in the previous chapter, relational practice is a respectful and reflexive approach to inquire into individuals' lived experiences and health care needs (Doane, 2002; Doane and Varcoe, 2015; Zou, 2016). In addition, it is the skilled action of respectful, compassionate, and authentically interested inquiry (Doane and Varcoe, 2015). The findings from the analysis of the parent interviews likewise suggest that what parents desire is not just good communication, but a sense of personal connection between themselves and healthcare professionals. Snaman et al. (2016) studied parents' perceptions of the communication patterns of healthcare providers while their children were dying. Overwhelmingly, the most commonly described positive communication strategy healthcare providers utilised was 'having a connection with the family' and not just treating them as a statistic.

Parents were often forced to re-examine pre-existing relationships and accommodate them in a changed form, for example, some described foregoing some friendships. The families' worlds became smaller as the children's care needs, appointments, and care coordination increasingly consumed their attention. Nonessential activities such as socialising were low priorities to parents who were stressed and sleep deprived. When social isolation became problematic for families through the loss of friends and inability to socialise, healthcare professionals helped fill this resultant void. Parents reported welcoming providers into their 'family' and having relationships that lasted sometimes for many years. The care, compassion and skill of certain healthcare professionals made parents feel valued. Davies, Baird and Gudmundsdottir (2013) explored bereaved fathers' perspectives of the care provided by healthcare professionals to the ill child and the family as a whole, both during the illness and throughout the dying process. The findings in the said study suggest that healthcare professionals require a baseline level of social adeptness, an ability to engage, interact, and develop relationships with families in crisis. Relationship difficulties with

healthcare professionals were an additional burden for parents to deal with, often leaving them feeling isolated and angry.

Whilst aspects of good service provision were highlighted, both parents and healthcare professionals identified that overall current services seemed under-resourced and insufficient to meet child and family needs. However, the perceived need to fight for services was more apparent in the parents' accounts. Lacking the physical presence of a support system, parents were aware that they shouldered the burden of care. Thus, fighting was perceived as essential in order to manage to care for their child. This mirrors the findings from other studies in which parents spoke about 'fighting the system' and their 'battle' for adequate service provision (Courtney et al., 2018; Dybvik et al., 2011; Price et al., 2012; Rallison and Raffin-Bouchal, 2013; Somanadhan and Larkin, 2016). Even with a support system in place parents struggled with severe emotional distress, significant fatigue, sleep impairment, difficulty in maintaining focus and sustaining hope, while living with constant uncertainty. While working with limited infrastructure and disorganisation resulted in healthcare professionals feeling pressurised and very concerned about the situation.

The results from the MPOC-20 offer complementary information with respect to experiences, including the experience of service provision. The results reveal both convergence and discrepancy or dissonance to the qualitative research findings. Firstly the results demonstrate that parents' perceptions regarding the care they were receiving were positive. Yet the qualitative research findings from the parents and the healthcare professionals reveal dissatisfaction regarding many aspects of care provision. However, parents and healthcare professionals talked about the quality of certain services. Parents were receiving services from a variety of service providers yet the focus when completing the MPOC-20 was on one service provider. The results demonstrate that the strongest aspect of service provision is related to the type of interaction and relationship parents have with healthcare professionals rather than any practical aspect of service provision. Parents and healthcare professionals emphasised the importance of this relationship. Yet parents' satisfaction with their relationship with healthcare professionals was extremely variable. Many parents talked about the bonds they had formed with healthcare professionals mirroring the findings of previously published studies (Bailey-Pearce et al., 2017; Carter, Edwards and Hunt, 2015; Hobson and Noyes, 2011). They likewise talked about positive and trusting relationships with healthcare professionals. Continuity of care was vital to parents, indicating that the trusting relationships that were built were important for parents. However, many of the parents were not connected with a consistent interdisciplinary team with whom ongoing, long-term relationships could be forged and nurtured.

This could explain the aforementioned finding. Additionally, this reflects the findings of previously mentioned studies (Rallison and Raffin-Bouchal, 2013; Somanadhan and Larkin, 2016).

The MPOC-20 domain Coordinated and Comprehensive Care were ranked high. Yet the parents indicated that care provision particularly at the time of diagnosis or recognition of a life-limiting condition was uncoordinated and unstructured. However, this aspect of care appeared to improve in time and in particular areas as indicated by both parents and healthcare professionals in this study. Additionally, the scores on this particular domain ranged considerably, reflecting different views with regard to whether the care received was in fact coordinated and comprehensive. This could be attributed to 'Different areas different services'. Likewise the theme 'Fragmented Health Care' emerged as a theme in a study undertaken to clarify the experiences and needs of families who had experienced a lethal fetal diagnosis. The said study was conducted in order to design responsive perinatal palliative care services (Cote-Arsenault and Denney-Koelsch, 2011). 'Fragmented Health Care' describes parents' disjointed and distant encounters with multiple providers at the time of diagnosis.

The MPOC-20 domains Providing General Information and Providing Specific Information were ranked lower than the other domains for parent satisfaction and correspond with qualitative data from parents. Yet the healthcare professionals suggested that communication and information exchange played an important role in supporting parents and families. However, all healthcare professionals identified the need for improved communication in order to enhance care for children with NMLLCs and their families. There were many examples of poor information exchange and breakdowns in communication between services. Collaboration within and between service providers was often fragmented. Participants in a previously mentioned study undertaken by Coad et al. (2015) reported that they had to retell their 'stories' many times to professionals because collaboration within and between service providers was often fragmented.

This study was not longitudinal in design so it cannot provide a measurement of change over time, however the parents' reflections on their experiences captured between one and sixteen years of their lives. The healthcare professionals who participated in this study were involved in the assessment and delivery of CPC in excess of five years. Both parents and healthcare professionals recognised changes within themselves that they attributed to their experience. Bronfenbrenner (1979) recognised that time and context resulted in changes to an individual's development. Parents in this study were exposed to changes in healthcare delivery because of policy change, government

priorities, and distribution of funding. The healthcare professionals were also exposed to these changes.

### **9.2.2 Access**

The qualitative and quantitative research findings from the parents were integrated previously regarding the issue of accessing services when caring for their child (see Chapter 6). Accordingly, minor discussion will take place at this juncture. As stated previously the qualitative and quantitative research findings from the parents were primarily consistent with regard to access. The analysis of the qualitative findings revealed that although parents entered the health system with a primary focus on dealing with their child's ill health, their attention became re-directed having to deal with the barriers in relation to accessing services and support. This finding was also reflected in the literature (Bradford et al., 2012; Noyes et al., 2013). 'Navigating the unknown territory' and 'Fighting for your child' came as an unexpected consequence of their healthcare engagement. In seeking care for their child, parents' expectations were that they would be embraced by healthcare professionals and presented with a clear pathway that facilitated appropriate assistance and support. The findings revealed that while parents focused on coping with the implications surrounding their child's condition they also had to deal with several other unanticipated barriers. Barriers consisted of firstly, trying to obtain appropriate care and secondly, finding appropriate supports (suitable services, understandable information and relevant resources) in order to cope with radical change and disruption to their lives. Barriers encountered encroached on precious time and attention taking the focus away from their child. Comparable findings have been reported in other studies (Dwybik et al., 2011; Price et al., 2012; Somanadhan and Larkin, 2016).

Accessing respite care was particularly problematic for parents in this study. The parents encountered multiple obstacles with regard to accessing respite. The findings from the demographic questionnaire provide additional information regarding the issue of accessing services. The most noteworthy finding was that some parents had access to two out-of-home respite services whereas others simply did not have access to any. This finding was corroborated by the healthcare professionals. The healthcare professionals highlighted inequalities in relation to the provision of out-of-home respite care to the extent that some families did not receive any whilst others, in similar circumstances in different areas received a lot. In some respects, some positive changes were gradually being evidenced over recent years by some parents in terms of services available. This finding was again substantiated by the healthcare professionals.

### **9.2.3 Decision-making regarding the care and management of children with NMLLCs**

The qualitative and quantitative research findings from the parents and the qualitative research findings from the healthcare professionals were chiefly consistent with regard to decision-making. The narratives revealed that parents of children with NMLLCs and healthcare professionals caring for this cohort of children have an imperative role to play in decision-making and regularly face decision-making dilemmas. This is consistent with previous published literature (Santoro and Bennett, 2018). Parents in particular revealed decisions that created incredible stress for themselves and their families. However, the provision of timely, accurate and appropriate information empowered parents to make decisions and thus reduced decisional conflict. This finding is supported in the literature (Boland et al., 2017). Indeed, a systematic review that specifically examined parental decision-making needs showed that parents require good quality information for example, available options, risks and benefits to make informed decisions on behalf of their child (Jackson, Cheater and Reid, 2008). Moreover, decisions were made easier when parents trusted healthcare professionals caring for their child and where parents were supported in their decisions. The importance of pre-existing relationships with healthcare professionals has been previously emphasised. Parents are more likely to discuss treatment decisions along a continuum (Boland et al., 2017; Larcher et al., 2015). However, parents were not always afforded the time or information that they required in order to make a decision. In addition, parents were not always offered treatment options or supported in their decisions.

The results from the DCS offer complementary information with respect to decision-making and therefore enhance knowledge and/or understanding of this phenomenon. Although parents reported a generally low level of decisional conflict in the DCS total scale and subscales the scores ranged from 0 (no decisional conflict) to scores exceeding 37.5 which is associated with decision delay or feeling unsure about implementation. This indicated that a small cohort of parents experienced high levels of decisional conflict. This is important as decisional conflict can be lowered with decision supporting interventions (O' Connor, 1993; Oshima and Emanuel, 2013). The results from the DCS make it possible to make observations and identify areas for enhancement regarding decision-making. It appeared that the attitudes and practices of individual healthcare professionals were the single most important modifiable factor that affected parental decision-making. An approach to decision-making where solutions emerge in the course of dialogue, disagreement and negotiation appeared to work best for both parents and indeed healthcare professionals.

Challenging healthcare professionals was difficult as parents were conscious of the effects of jeopardising the relationship they had with them.

Discrepancies were also noted between the narratives. The perceived need to make complex decisions were more apparent in the parents' accounts. Parents repeatedly spoke about the need to engage with decisions that regularly included ordinary, day-to-day queries not connected to critical or end-of-life decisions. As previously outlined (see Chapter 3) Koch and Jones (2018) suggest that these parents engage with decision-making that often includes considering mundane, everyday questions not related to end-of-life conversations or processing a new illness status or diagnosis. The parents revealed that participation in decision-making related to the presence or absence of apparent choice and occurred in circumstances where the child's condition dictated a particular step to be taken. This finding was also mirrored in the literature (Nicholson, 2012). However, this view was rarely acknowledged by healthcare professionals. The healthcare professionals appeared to focus their attention on end-of-life care conversations and decisions. Historically, CPC has focused on end-of-life decision-making (Koch and Jones, 2018). Research relating to decision-making for children with life-limiting conditions is largely focused on end-of-life care (Beecham et al., 2017; Bluebond-Langer et al., 2017). As the field continues to grow and children with life-limiting conditions continue to live longer, healthcare professionals should acknowledge the need for continued assessment, treatment, and referral to address the needs of parents of children who may have conditions that span decades.

It is acknowledged that decisions surrounding CPC often occur in contexts of uncertainty and ethical dilemmas (Hoell, 2017). It is also important to acknowledge that there is rarely one overarching ethical principle or value that can be used to solve an ethical problem. Parents hold personal ethical values that must be recognised (Glasper, 2017). Like the parents, the healthcare professionals believe that decisions should be made in the best interest of the child (Popejoy et al., 2017). However, this is often difficult to ascertain particularly in the early stage of the care trajectory. Additionally, there are times when parents and healthcare professionals are required to make decisions in a dynamic and time-sensitive manner. The healthcare professionals have greater medical knowledge and experience and their opinion is likely broader and more objective in times of crises. In addition, the healthcare professionals often consult with other professionals and reach recommendations based on a number of factors. Parents and healthcare professionals in the present study were mindful of this. Parents' preferences for delegating decision-making control to the healthcare professionals were associated with their perceptions of a high degree of urgency.

This mirrors the findings of a study undertaken with parents of infants in neonatal intensive care (Weiss et al., 2016). Parents in the present study described the need to have come to a 'place of acceptance' in order to be able to take part in decision-making. In contrast, preferences for greater parental control were associated with high parental knowledge or personal experience with the decision.

#### **9.2.4 Summary of the integrative analysis**

Integrating the findings to emerge across the entire study was important as it enabled the identification of convergent, complementary, silent and discrepant or dissonant findings. This generated a deeper insight than separate analyses. The qualitative and quantitative research findings from the parents and the qualitative research findings from the healthcare professionals were chiefly consistent across the entire study.

### **9.3 The original contribution of the present study**

This study's unique contribution is made through providing new knowledge relating to the experience of caring for children with NMLLCs on the island of Ireland from the perspective of parents and healthcare professionals. Regardless of some notable differences in CPC service provision between Ireland and Northern Ireland, for example, the greater level and availability of hospice services for children with life-limiting conditions in Northern Ireland than for children in Ireland, parents and healthcare professionals in both jurisdictions shared similar experiences. This could be attributed to the similarities between the strategic policies and guidelines between Ireland and Northern Ireland (DoHC, 2009; DHSSPS, 2014; DoH, 2016). The findings provide new insights regarding clinical expertise and the concept of the healthcare professional as expert. Benner's model of skill acquisition is widely used in nursing practice, research and education. Benner (1982) detailed the acquisition of expertise and proposed five possible expertise levels: novice, advanced beginner, competent, proficient, and expert. The healthcare professionals in this study evidently progressed through the stages to become the expert. They had a comprehensive knowledge base. They also had an intuitive grasp of each situation and their performance became fluid and flexible and highly proficient. However, the concepts of relational practice, family-centred care and liminality were also apparent within the healthcare professionals accounts' and appeared to give rise to the very clinically competent expert within this specialised field of practice. It is evident that these core concepts need to be recognised and integrated into existing models. As previously reported a structural failure to recognise liminality can be detrimental to parents and families (Jordan, Price and Prior, 2015). Additionally, failure to incorporate family-centred care can result in

multiple adverse consequences including difficulties with communication and dissatisfaction with care. Furthermore the findings from this study suggest that delayed relational practice can cause unnecessary stress for parents and families and can increase the risk of further harm. This finding is supported in the literature (Zou, 2016). To the author's knowledge, no previous research has utilised mixed methodology to explore the experience of caring for children with NMLLCs on the island of Ireland from the perspective of parents and healthcare professionals.

#### **9.4 Strengths and limitations of the study**

In this section the overarching strengths and limitations of this mixed methods study are discussed. The findings from this study provide rich and nuanced insights into what it means to be a parent of a child with a NMLLC. Children with NMLLCs represent a group with conditions which, on their own, are few in number, lack visibility and whose needs are complex, difficult to define but very debilitating (Adams, Miller and Grady, 2016; Nageswaran, Hurst and Radulovic, 2018). Due to the small numbers of children and the rarity of many of the life-limiting conditions parents often lack a collective voice. Importantly parents of children from all four categories of childhood life-limiting conditions were included in this study. However, parents of children with cancer were excluded as both the illness trajectory and CPC services differ significantly for these children (Bergstrasser, 2018; Price, 2012). The findings also provide insights regarding how parents navigate the complex process of care provision, including accessing services and support on the island of Ireland. The findings likewise provide an insight relating to how healthcare professionals experience the delivery of care required by children and their families. The healthcare professionals in this study had a variety of roles in the delivery of CPC service provision, including a paediatrician, a social worker, a neonatologist, a clinical psychologist and an Outreach nurse. Previous studies almost exclusively included nurses and doctors, with very little involvement of other relevant healthcare professionals (Baverstock and Finlay, 2008; Pontin and Lewis, 2008; Quinn and Bailey, 2011). It was important to include a variety of healthcare professionals in this study given the multidisciplinary nature of CPC (Basu and Swil, 2018; Benini et al., 2008; Doug et al., 2011; Postier, Catrine and Remke, 2018). It was critical to include the perspective of parents and healthcare professionals, thus ensuring the most comprehensive picture of palliative care practice. Evidence from this study and from various fields suggest that what is important to parents and families can be different to what is important to healthcare professionals (Haywood, Collin and Crawley, 2014; Kirwan et al., 2009; Michener, 2011; Serrano-Anguilal et al., 2009). The mixed methods design of the present study denotes a key strength of the overall study as it enabled a comprehensive exploration of the experience of caring for children with NMLLCs including, accessing services and decision-making surrounding the child's

care; a topic that had previously received inadequate attention in CPC literature. In spite of the strengths of the present study, a number of limitations also existed. Notably fathers were underrepresented in the present study. Despite being open to both parents, the sample encompassed mostly mothers, given very few fathers volunteered to partake. Thus, when interpreting the findings, it is important to note the over-representation of mothers which may have obscured gender perspective differences. Despite efforts to recruit participation from a diverse range of respondents, there was little diversity among parents in this sample. The sample was homogenous in terms of ethnic profile with all parents of Irish ethnic background. Whilst the limitations of the present study existed, because of the very difficult nature of accessing parents, it is posited that rather than being actual weaknesses and substantially detracting from the value of the findings, the limitations are issues that researchers could aim to address within future studies. This will be discussed in the section on future research.

## **9.5 Implications for practice, future research, education and policy**

The experiences shared by parents and healthcare professionals point to some important practice, research, education and policy considerations and are largely supported by previous and relevant healthcare research. Parents in this study illuminated the everyday experience of living with a child with a NMLLC. Such experiences raise important considerations for improvement in supports, with implications for both practice-and policy-level initiatives.

### **9.5.1 Practice implications**

The findings from this study bring to light many pragmatic considerations for healthcare professionals and service providers. The findings support the use of the FMSF, Paterson's Shifting Perspectives Model of Chronic Illness and Bronfenbrenner's Ecological Theory for this group of parents and families. The use of the FMSF and Paterson's Shifting Perspectives Model of Chronic Illness in evaluating family management can aid healthcare professionals in planning tailored interventions that maximise child and family outcomes. However, it is evident that consideration should likewise be given to other influences such as the system that impact on both the child and family therein. The context and environment shapes access and experiences of healthcare. Bronfenbrenner's theory reminds healthcare professionals that sometimes they need to advocate for changes in organisations and systems to best support children with NMLLCs and their families. Consequently the amalgamation of the FMSF, Paterson's Shifting Perspectives Model of Chronic Illness and Bronfenbrenner's Ecological Theory are appropriate. Despite developing a changed, valued perspective regarding what is meaningful in life, the significant isolation and exclusion from

the community and the workforce as a result of accepting the caregiver role was profound for parents. In the background of diminished friendships and social networks for a majority of parents, a peer mentoring or online service which is adequately resourced and facilitated by healthcare professionals may help to build community and reduce isolation felt by parents. Sharing their unique expertise developed through caregiving with others may additionally enable parents to derive ongoing meaning from their experience. The findings suggest a discrepancy between services offered, current standards of care and recognition of families' difficulty or sometimes inability to access and utilise services and supports outside the home. The findings from this research demonstrate that the MPOC-20 is a useful tool in enabling service providers to identify areas of improvement and move towards services that are more family-centred. Importantly the findings from this study revealed that family-centred care was not always transparent within organisations particularly in hospital environments. Indeed, adherence to the principles of family-centred care appeared to be at the discretion of individual healthcare professionals. In a previously mentioned study fathers identified healthcare professionals who engaged in family-centred care as exceptional (Davies, Baird and Gudmundsdottir, 2013). The fact that fathers identified healthcare professionals who engaged in family-centred care as exceptional indicates that this level of care is not being consistently delivered. Engaging fully in family-centred care may also require a re-evaluation of the concept of professional boundaries (Jankowski, 2014). In almost every discipline, providers are taught to maintain strict boundaries and to avoid engagement in the very types of relationships that the parents valued most. Despite instruction to the contrary, many healthcare professionals do engage in some level of boundary-crossing, calling into question the relevancy of such boundaries. The findings support the need to measure decisional conflict for this cohort of parents and families. Given the harmful effects of unresolved decisional conflict, the findings suggest that a significant number of parents would benefit greatly from decision aids (Delany et al., 2017; Xafis et al., 2015), decision coaching (O' Connor, Stacey and Legare, 2008) or from their healthcare providers being trained in shared decision-making. Evidence based interventions should be regularly evaluated (Boland et al., 2017). The FMSF and Paterson's Shifting Perspectives Model of Chronic Illness is fitting to help understand how families approach decision-making. For example, in the interest of the family and maintaining normalcy, parents may have a tendency to resist decisions that are perceived as triggering intrusion of the condition in everyday life.

### **9.5.2 Future research**

Additional research is essential to contribute to the optimal care and well-being of these children and their families. Future research should endeavour to further elucidate the perspectives of

fathers. In addition, the needs of parents of children with NMLLCs from a multicultural and cross cultural perspective should be explored. Also, other professional groups outside of the healthcare sectors such as priests/chaplains and teachers may bring new insights and perspectives. The experience of siblings of children with NMLLCs in an Irish context should also be sought. The voice of the child has been rarely represented in the current literature base. However, the inclusion of children and young people present challenges given the nature of the conditions concerned. An ethnographic study may be appropriate in the future. Although there have been strides in end-of-life decision-making, further investigation and research is needed in this field.

### **9.5.3 Education**

Relational practice emerged as important to the healthcare professionals' ability to be with a family and provide support throughout the care trajectory from diagnosis through to end-of-life care and following the death of the child. Relational learning approaches strive for healthcare professionals to achieve high levels of discipline-specific competence along with the communicative and relational skills necessary for establishing meaningful connections (Konrad and Browning, 2012). Relational practice, in conjunction with family-centred care and liminality should be taught as foundational concepts in CPC curricula. Additionally, purposeful connections need to be delineated between the theory of these concepts and their application into practice. Communicating with parents and families, particularly at the time of diagnosis and in relation to end-of-life care and decision-making is extremely challenging for healthcare professionals. The healthcare professionals need to engage parents and families in critical, challenging conversations. The skills required, especially techniques for effective communication in emotionally charged situations, need to formally enter the curricula for healthcare professionals who are caring for children with life-limiting conditions. This is supported in previous CPC research studies which likewise suggest that skills, particularly techniques for effective communication in highly emotional situations, need to enter the curricula in both theory and practice (Kenny et al., 2016; MacDonnell-Yilmaz, 2015). By enhancing the communication skills of healthcare professionals it will enable them to engage in critical conversations with the family. As interdisciplinary practice is central to CPC, interdisciplinary education should be offered to healthcare professionals. A number of healthcare professionals in this study described initially feeling overwhelmed and fearful in caring for children and families. Interestingly, some of the same fears of providing adequate support for the child and family are reported in the literature across disciplines (Basu and Swil, 2018; Quinn and Bailey, 2011; Reid, 2013).

#### **9.5.4 Policy implications**

The findings from this study raise a number of policy implications that may be embarked upon in an effort to facilitate a greater level of support for parents/families and healthcare professionals caring for children with NMLLCs. Collaboration is essential in providing a quality service to these children and families. Services should be organised in a manner which are more appropriate to facilitate the specific needs of these children and families. Parents often perceived that the child's needs could be best supported in the family home. Thus, more readily available in-home psychological, counselling and respite services are necessary in conjunction with out-of-home options, to enable improved access to supports. Care should be adaptable to the individual and changing needs of the child and his or her family. High quality care should be provided regardless of location. Families need the reassurance of having support particularly at end-of-life. Services and support should be available 24 hours a day. The development of a national database of children with life-limiting conditions is critical for future planning. The suggestion of supplementing the existing categories of life-limiting and life-threatening conditions by identifying precise diagnoses should be considered. Clear guidance surrounding clinical governance is required for healthcare professionals working in this area of practice.

#### **9.6 Concluding comments**

The findings from this research suggest for the first time that there is a relationship between parents and healthcare professional experiences of caring for children with NMLLCs including their experience of CPC service provision and decision-making, findings which have significant clinical implications. While much focus is placed on the clinical care needs of children with life-limiting conditions, future work will need to consider the implementation of carefully planned support services and other societal initiatives which recognise and seek to alleviate the broad impacts on parents. Although the provision of care closer to home for such children is a policy objective, internationally integration of health services is insufficient with wide variation in systems of care for these children and their families (Peter et al., 2011). This is also demonstrated in the present study. Yet parents' views regarding care generally match current CPC policy aspirations.

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## Appendix A: Letter of invitation: parents.

Dear parent,

I am a Children's Nurse. Currently, I am conducting a research study as part of my PhD in Dublin City University. My research is under the guidance of two university supervisors, Dr. Gemma Kiernan and Professor Jayne Price.

The title of the study is: **'An exploration of the experience of caring for children with non-malignant life-limiting conditions including, accessing services and decision-making surrounding the child's care.'**

I am contacting you as you are a parent of a child who accesses services provided by (named service provider). I would appreciate if you could partake in the research as your experiences are very relevant to this study. Enclosed is information about the study. Before you decide whether you wish to partake or not, I would like if you could take the time to read the information.

If you would like any further information or if anything in the documents is unclear, please contact me at the following telephone number 00 353 87 3812250 or alternatively by email at [fiona.hurley4@mail.dcu.ie](mailto:fiona.hurley4@mail.dcu.ie) and I will be happy to discuss the study further with you.

Kind regards, Fiona Hurley

-----  
Next steps:

1. Please read the enclosed Information and Consent form.
2. Decide whether you would like to partake in the study.
3. Complete the Expression of Interest slip if you are interested in partaking and return it to me.
4. Please feel free to contact me with any queries. I look forward to further contact with you in relation to this research study.

## **Appendix B: Information regarding the study: parents.**

**Title of the study is:** 'An exploration of the experience of caring for children with non-malignant life-limiting conditions including, accessing services and decision-making surrounding the child's care'.

### **Investigators**

Ms. Fiona Hurley. School of Nursing and Human Sciences. Dublin City University.  
Telephone: +353 87 3812250. Email: [fiona.hurley4@mail.dcu.ie](mailto:fiona.hurley4@mail.dcu.ie)

Dr. Gemma Kiernan. School of Nursing and Human Sciences. Dublin City University.  
Telephone: +353 1 7008542 Email: [gemma.kiernan@dcu.ie](mailto:gemma.kiernan@dcu.ie)

Professor Jayne Price. School of Nursing, Kingston University and St. George's University of London.  
Telephone: + 44 (0)20 8417 5703 Email: [j.e.price@sgul.kingston.ac.uk](mailto:j.e.price@sgul.kingston.ac.uk)

### **What is the research about?**

This research is about exploring the experiences of parents caring for a child with a non-malignant life-limiting condition, including their views of accessing services. It also examines decision-making regarding the care and management of children with non-malignant life-limiting conditions. Additionally, it examines the experiences of healthcare professionals caring for this group of children.

### **Why is the research being conducted?**

As a group, children with non-malignant life-limiting conditions have been identified as benefiting from a comprehensive palliative care service. However, despite the fact that these children constitute the largest proportion of children requiring palliative care, services are often fragmented. Additionally, there is a lack of information on the experiences of parents caring for a child with a NMLLC, including their views of accessing services and decision-making surrounding care. There is also a lack of information on the experience of healthcare professionals caring for this group of children. This research will incorporate the perspectives of parents and healthcare professionals, thus ensuring the most comprehensive picture of current palliative practice, including access to services, decision-making and service provision. Ultimately, the findings from the research will be used to make recommendations for a model of service provision.

### **Time/ what is involved in the research?**

Parents will be invited to participate in an interview. The interview will be **audio** recorded with permission. A number of questions will be asked regarding parents personal experiences of caring for their child, including their experiences of palliative care services. The interview will not exceed 90 minutes. Parents will also be asked to complete a demographic questionnaire and two measurement scales at the end of the interview, which will take approximately 20 minutes each to complete. The demographic questionnaire will be used to gather background information on the family makeup and other factors such as where the family live which may impact on access and experience of services.

### **How will their privacy be protected?**

Every effort will be made by the researcher to protect the identity of the participants. All data returned in the questionnaires will be treated as confidential. Data will be transferred to a secure, password protected laptop which will be in the sole custody of the researcher throughout the study. When not in use the laptop will be stored in a locked cabinet in the researcher's office. All identifying information will be removed from the interview transcripts, including names, locations and named services and institutions. However, in highly specialised areas the identity of

participants can sometimes be logically deduced. For example, children with non-malignant life-limiting conditions have often rare conditions, thus it is possible that a child/parent may be easily identified if the child's diagnosis is specifically recorded. Every effort will be made by the researcher to remove such opportunities for others to deduce the child or parent's identity from the data. The researcher will remove all potential identifiers from any report, publication or presentation.

### **How will the data be used and subsequently disposed of?**

The findings will be analysed and published in a report. The findings will also be published in appropriate Journals and presented at suitable conferences. All data records will be retained for a period of five years following completion of the study. This is required by Dublin City University. Additionally, once the PhD is awarded, the anonymised information collected will be stored carefully so that other researchers can use it in years to come. This will only be available to the researchers after a period of two years post award of the PhD.

### **What are the legal limitations to data confidentiality?**

The researcher is required to disclose information if matters relating to the safety of a child are raised during the interview. The researcher will adhere to appropriate guidelines with regard to this. Should this situation arise the researcher will stop the interview and will discuss the action that needs to be taken with the parent.

### **Benefits of taking part in the study**

All of the participants will have an opportunity to voice their opinion on service provision for children with non-malignant life-limiting conditions. It is hoped that the findings will contribute towards improving the quality of care for children and their parents/families.

### **Risks of taking part in this research study**

There are no risks associated with taking part in this research. Should any parent display signs of discomfort during an interview, the interview will be stopped and the parent will be given the opportunity to withdraw from the study. If this situation arises, the researcher will contact the parent within 48 hours of withdrawing from the study to check on their wellbeing and will seek their permission to contact a healthcare professional for ongoing support if they remain distressed.

### **Can participants change their mind at any stage and withdraw from the study?**

Taking part in the study is a matter of choice. Parents who decide not to participate will not experience any negative impact on the care they or their child receives. Written consent will be obtained, from parents who agree to participate, however participants' can withdraw from the study at any time without the need to give a reason for their decision.

### **Contact details**

For more information about the study please contact Fiona Hurley.

Email: [fiona.hurley4@mail.dcu.ie](mailto:fiona.hurley4@mail.dcu.ie)

***If participants have concerns or complaints about the process involved in this study and wish to contact an independent person please contact \_\_\_\_\_ Telephone \_\_\_\_\_***

*Please note that complaints or concerns that are not satisfactorily resolved will be progressed in accordance with Dublin City University procedures.*

**Appendix C: Expression of Interest Slip: parents.**

Parent(s) Name: .....

Child's Name: .....

Diagnosis (if known): .....

Home Address: .....

Telephone Number: .....

Best time to call: .....

*Thank you*

## Appendix D: Consent for interview: parents.

**Title of the study:** 'An exploration of the experience of caring for children with non-malignant life-limiting conditions including, accessing services and decision-making surrounding the child's care'.

### Investigators

Ms. Fiona Hurley. School of Nursing and Human Sciences. Dublin City University.

Dr. Gemma Kiernan. School of Nursing and Human Sciences. Dublin City University.

Professor Jayne Price. School of Nursing, Kingston University and St. George's University of London.

### Purpose of the Study

The purpose of this research is to explore the experience of caring for children with NMLLCs including, the experience of accessing services and decision-making surrounding the child's care.

### Consent

Written consent will be obtained, however participants' can withdraw from the study at any time without the need to give a reason for their decision.

### Please indicate your consent to be interviewed for this study by completing this form.

I have had a chance to discuss and ask questions about this study.

Yes                  No

<input type="checkbox"/>	<input type="checkbox"/>
--------------------------	--------------------------

I understand what the study is about.

Yes                  No

<input type="checkbox"/>	<input type="checkbox"/>
--------------------------	--------------------------

I understand that this interview will be audio-recorded.

Yes                  No

<input type="checkbox"/>	<input type="checkbox"/>
--------------------------	--------------------------

I know that the privacy of information I provide can only be guaranteed within the limitations of the law.

Yes                  No

<input type="checkbox"/>	<input type="checkbox"/>
--------------------------	--------------------------

I know that, in order to minimise the chance of identifying interviewees, the researchers will not attribute comments to individual participants.

Yes                  No

<input type="checkbox"/>	<input type="checkbox"/>
--------------------------	--------------------------

I am aware that absolute confidentiality cannot be guaranteed as this research is highly specialised. The number of children living with non-malignant life-limiting conditions in Ireland are small and thus it may be possible for the identity of a participant to be deduced.

Yes                  No

<input type="checkbox"/>	<input type="checkbox"/>
--------------------------	--------------------------

I know that if I wish, I am free to stop this interview at any time, or withdraw from the study, without having to explain my decision, or suffer any adverse effects.

Yes                  No

<input type="checkbox"/>	<input type="checkbox"/>
--------------------------	--------------------------

I am aware that anonymised information collected will be stored carefully in an archive so that other researchers can use it in years to come.

Yes	No
<input type="checkbox"/>	<input type="checkbox"/>

My questions and concerns have been answered and I have retained a copy of this consent form. Therefore, I consent to take part in this research project.

\_\_\_\_\_  
Signature of participant

Date \_\_\_\_\_

\_\_\_\_\_  
Name in block capitals

\_\_\_\_\_  
Signature of researcher

Date \_\_\_\_\_

## Appendix E: Letter of invitation: healthcare professionals.

Dear healthcare professional,

I am a Registered Children's Nurse. Currently, I am conducting a research study as part of my PhD in Dublin City University. My research is under the guidance of two university supervisors, Dr. Gemma Kiernan and Professor Jayne Price. The title of the study is: **'An exploration of the experience of caring for children with non-malignant life-limiting conditions including, accessing services and decision-making surrounding the child's care'**.

I am contacting you as you are a healthcare professional involved in the assessment and provision of care to this cohort of children. I would appreciate if you could partake in the research as your experiences are very relevant to this study. Enclosed is information about the study. Before you decide whether you wish to partake or not, I would like if you could take the time to read the information.

If you would like any further information or if anything in the documents is unclear, please contact me at the following telephone number 00 353 87 3812250 or alternatively by e mail [fiona.hurley4@mail.dcu.ie](mailto:fiona.hurley4@mail.dcu.ie) and I will be happy to discuss the study further with you.

Kind regards,

-----  
Fiona Hurley

Next steps:

1. Please read the enclosed information sheet and consent form.
2. Decide whether you would like to partake in the study.
3. Complete the Expression of Interest slip if you are interested in partaking and return it to me.
4. Please feel free to contact me with any queries. I look forward to further contact with you in relation to this research study.

## **Appendix F: Information sheet regarding the study: healthcare professionals.**

**Title of the study:** ‘An exploration of the experience of caring for children with non-malignant life-limiting conditions including, accessing services and decision-making surrounding the child’s care’.

### **Investigators**

Ms. Fiona Hurley. School of Nursing and Human Sciences. Dublin City University.  
Telephone: 00 353 87 3812250. Email: [fiona.hurley4@mail.dcu.ie](mailto:fiona.hurley4@mail.dcu.ie)

Dr. Gemma Kiernan. School of Nursing and Human Sciences. Dublin City University.  
Telephone: +353 1 7008542. Email: [gemma.kiernan@dcu.ie](mailto:gemma.kiernan@dcu.ie)

Professor Jayne Price. School of Nursing, Kingston University and St. George’s University of London.  
Telephone: + 44 (0)20 8417 5703. Email: [j.e.price@sgul.kingston.ac.uk](mailto:j.e.price@sgul.kingston.ac.uk)

### **What is the research about?**

This research is about exploring the experiences of parents caring for a child with a non-malignant life-limiting condition, including their views of accessing services. It also examines decision-making regarding the care and management of children with non-malignant life-limiting conditions. Additionally, it examines the experiences of healthcare professionals caring for this cohort of children.

### **Why is the research being conducted?**

As a group, children with non-malignant life-limiting conditions have been identified as benefiting from a comprehensive palliative care service. However, despite the fact that these children constitute the largest proportion of children requiring palliative care, services are often fragmented. Additionally, there is a lack of information on the experiences of parents caring for a child with a NMLLC, including their views of accessing services and decision-making surrounding care. There is also a lack of information on the experience of healthcare professionals caring for this cohort of children. This research will incorporate the perspectives of parents and healthcare professionals, thus ensuring the most comprehensive picture of current palliative practice, including access to services, decision-making and service provision. Ultimately, the findings from the research will be used to make recommendations for a model of service provision.

### **Time/ what is involved in the research?**

Healthcare professionals will be invited to participate in an interview. The interview will be **audio** recorded with permission. Open-ended questions will be used to explore healthcare professionals’ experiences of caring for children with non-malignant life-limiting conditions. The interview will not exceed 90 minutes.

### **How will their privacy be protected?**

Every effort will be made by the researcher to protect the identity of the participants. Data will be transferred to a secure, password protected laptop which will be in the sole custody of the researcher throughout the study. When not in use the laptop will be stored in a locked cabinet in the researcher’s office. All identifying information will be removed from the interview transcripts, including names, locations and named services and institutions. Access to computerised data will be available only to the researcher, transcriber and her academic supervisors during the course of the study. The researcher will remove all potential identifiers from any report, publication or presentation.

### **How will the data be used and subsequently disposed of?**

The findings will be analysed and published in a report. The findings will also be published in appropriate Journals and presented at suitable conferences. 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. Additionally, once the PhD is awarded, the anonymised information collected will be stored carefully in an archive so that other researchers can use it in years to come. This will only be available to the researchers after a period of two years post award of the PhD.

### **What are the legal limitations to data confidentiality?**

The researcher is required to disclose information if matters relating to the safety of a child are raised during the interview. The researcher will adhere to appropriate guidelines with regard to this. Should this situation arise the researcher will stop the interview and will discuss the action that needs to be taken.

### **Benefits of taking part in the study**

All of the participants will have an opportunity to voice their opinion on service provision for children with non-malignant life-limiting conditions. It is hoped that the findings will contribute towards improving the quality of care for children and their parents/families.

### **Risks of taking part in this research study**

There are no risks associated with taking part in this research study. Should any healthcare professional display signs of discomfort or distress during an interview, the interview will be stopped and the healthcare professional will be given the opportunity to withdraw from the study.

### **Can participants change their mind at any stage and withdraw from the study?**

Written consent will be obtained, however participants' can withdraw from the study at any time without recrimination.

### **Contact details**

For more information about the study please contact Fiona Hurley.  
Email: [fiona.hurley4@mail.dcu.ie](mailto:fiona.hurley4@mail.dcu.ie)

***If participants have concerns or complaints about the process involved in this study and wish to contact an independent person please contact \_\_\_\_\_ Telephone: \_\_\_\_\_***

*Please note that complaints or concerns that are not satisfactorily resolved will be progressed in accordance with Dublin City University procedures.*

**Appendix G: Expression of Interest Slip: healthcare professionals.**

Name: .....

Name of organisation:.....

Professional role: .....

Number of years' experience in this role: .....

Email address: .....

Telephone Number: .....

Best time to call: .....

*Thank you*

## Appendix H: Consent for interview: healthcare professionals.

**Title of the study:** 'An exploration of the experience of caring for children with non-malignant life-limiting conditions including, accessing services and decision-making surrounding the child's care'.

### Investigators

Ms. Fiona Hurley. School of Nursing and Human Sciences. Dublin City University.  
Telephone: 00 353 87 3812250. Email: [fiona.hurley4@mail.dcu.ie](mailto:fiona.hurley4@mail.dcu.ie)

Dr. Gemma Kiernan. School of Nursing and Human Sciences. Dublin City University.  
Telephone: +353 1 7008542. Email: [gemma.kiernan@dcu.ie](mailto:gemma.kiernan@dcu.ie)

Professor Jayne Price. School of Nursing, Kingston University and St. George's University of London.  
Telephone: + 44 (0)20 8417 5703. Email: [j.e.price@sgul.kingston.ac.uk](mailto:j.e.price@sgul.kingston.ac.uk)

### Purpose of the Study

The purpose of this research is to explore the experience of caring for children with NMLLCs including, the experience of accessing services and decision-making surrounding the child's care.

### Consent

Written consent will be obtained, however participants' can withdraw from the study at any time without recrimination.

### Please indicate your consent to be interviewed for this study by completing this form.

I have had a chance to discuss and ask questions about this study.

Yes	No
<input type="checkbox"/>	<input type="checkbox"/>

I understand what the study is about.

Yes	No
<input type="checkbox"/>	<input type="checkbox"/>

I understand that this interview will be audio-recorded.

Yes	No
<input type="checkbox"/>	<input type="checkbox"/>

I know that the privacy of information I provide can only be guaranteed within the limitations of the law.

Yes	No
<input type="checkbox"/>	<input type="checkbox"/>

I know that, in order to minimise the chance of identifying interviewees, the researchers will not attribute comments to individual participants.

Yes	No
<input type="checkbox"/>	<input type="checkbox"/>

I am aware that absolute confidentiality cannot be guaranteed as this research is highly specialised. The number of healthcare professionals caring for children with non-malignant life-limiting conditions in Ireland are small thus it may be possible for the identity of a participant to be deduced.

Yes	No
<input type="checkbox"/>	<input type="checkbox"/>

I know that if I wish, I am free to stop this interview at any time, or withdraw from the study, without having to explain my decision, or suffer any adverse effects.

Yes	No
<input type="checkbox"/>	<input type="checkbox"/>

I am aware that the anonymised information collected from the interviews will be stored carefully in an archive so that other researchers can use it in years to come.

Yes	No
<input type="checkbox"/>	<input type="checkbox"/>

My questions and concerns have been answered and I have retained a copy of this consent form. Therefore, I consent to take part in this research project.

\_\_\_\_\_  
Signature of participant

Date \_\_\_\_\_

\_\_\_\_\_  
Name in block capitals

\_\_\_\_\_  
Signature of researcher

Date \_\_\_\_\_

## Appendix I: Interview Guide: parents.

The interview will begin by thanking the parent for giving their time. The researcher will initiate general conversation to help the parent feel at ease and to encourage mutual sharing of information. Before the interview commences the researcher will ask for permission to turn the audio recorder on. The purpose of the interview will be reiterated and the researcher will go through the consent form. The parent will be reminded that the interview may be paused or stopped at any time.

The interview will focus on the following topics:

- the experiences of parents caring for their child
- parents views of accessing services
- Decision-making

Interview topic.	Interview guide questions.
Experiences.	<ul style="list-style-type: none"> <li>• Could I begin by asking you to tell me about (child’s name)?</li> </ul> <p>The subsequent questions may be covered by the parent.</p> <ul style="list-style-type: none"> <li>• Could you tell me about the time you first had concerns regarding your child?</li> <li>• Could you tell me what it was like for you at that time?</li> <li>• How has it impacted on you and your family? <b>Prompts ...</b> daily life/family life. ???Giving up work...how did you come to that decision?</li> </ul>
Experiences of palliative care services including issues of access.	<p>Could you tell me about the services your child receives?</p> <p><b>Prompts...</b>Home care, hospital care, respite. ???Transition if appropriate</p> <p>Accessibility of services.</p>
Decision-making.	<p>Since (child’s name) was diagnosed have you had to make a lot of decisions about his/her health care? <b>Prompts.....</b>how has this been?</p> <p>Who supports you in your decisions?</p>

Finally the parent will be asked to reflect on the healthcare professionals that they regularly engage with.....

Could you tell me about the healthcare professionals that are most significant in (child’s name) life?

Who is your key contact in terms of healthcare professionals at present?

## Appendix J: Interview Guide: healthcare professionals.

The interview will begin by thanking the healthcare professional for giving up their time. The researcher will initiate general conversation to help the healthcare professional feel at ease and to encourage mutual sharing of information. Before the interview commences the researcher will ask for permission to turn the audio recorder on. The purpose of the interview will be reiterated and the researcher will go through the consent form. The healthcare professional will be reminded that the interview may be paused or stopped at any time. The interview will focus on the following topics:

- The experiences of healthcare professionals caring for children with NMLLC s
- Decision-making.

Interview topic.	Interview guide questions.
Experiences	<ul style="list-style-type: none"> <li>• Could I begin by asking you to tell me about your professional role in caring for children with NMLLCs?</li> </ul> <p>The subsequent questions may be covered by the healthcare professional.</p> <ul style="list-style-type: none"> <li>• How long have you worked in this area?</li> <li>• Could you tell me about your early experiences?</li> </ul> <p><b>Prompts</b>.....education and its influence....emotional impact of caring for this cohort of children .....challenges that families face in caring for children with NMLLCs</p> <ul style="list-style-type: none"> <li>• Could you tell me about services that children and their families’ access? .....Service distribution...equity ...challenges in caring</li> </ul>
Decision-making.	<p>The healthcare professional will be asked to reflect on decisions they have made in relation to children with NMLLCs and their families.</p> <p>Examples of questions that a healthcare professional may be asked are .....</p> <p>What influences your decision to refer a child to palliative care? (If it is applicable in their role).</p> <p>What are the most challenging decisions you face as a healthcare professional?</p> <p>Who do you think should be the ultimate decision-maker with regard to a child receiving palliative care?</p> <p>To what extent should the child/family have a voice in any treatment/palliative care decisions?</p>

## Appendix K: Decisional Conflict Scale.

Considering the option you prefer, please answer the following questions

	<b>Strongly Agree</b>	<b>Agree</b>	<b>Neither Agree nor Disagree</b>	<b>Disagree</b>	<b>Strongly Disagree</b>
	(0)	(1)	(2)	(3)	(4)
1. I know which options are available to me.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. I know the benefits of each option.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. I know the risks and side effects of each option.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. I am clear about which benefits matter most to me.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
5. I am clear about which risks and side effects matter most to me.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
6. I am clear about which is more important to me (the benefits or the risks and side effects).	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
7. I have enough support from others to make a choice.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
8. I am choosing without pressure from others.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
9. I have enough advice to make a choice.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
10. I am clear about the best choice for me.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
11. I feel sure about what to choose.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
12. This decision is easy for me to make.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
13. I feel I have made an informed choice.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
14. My decision shows what is important to me.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
15. I expect to stick with my decision.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
16. I am satisfied with my decision.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

## Appendix L: The Measure of Processes of Care-20 - (MPOC-20).

We would like to understand and measure the experiences of parents who have a child with a disability. In particular we wish to know about your perceptions of the care you have been receiving over the past year from the health care organisation that provides services to your child. This may be your local children=s treatment (rehabilitation) centre, your community care access centre, or another organisation. The care that you and your child receive from this organisation may bring you into contact with many individuals. The questions on this form are grouped by who these contacts are, as described below.

**PEOPLE:** refers to those individuals who work directly with you or your child. These **may include** psychologists, therapists, social workers, doctors, teachers, etc.

**ORGANISATION:** refers to all staff from the health care organisation, whether involved directly with your child or not. In addition to health care people they **may include** support staff such as office staff, housekeepers, administrative personnel, etc.

The questions are based on what parents, like yourself, have told us about the way care is sometimes offered. We are interested in your personal thoughts and would appreciate your completing this questionnaire on your own without discussing it with anyone.

For each question, please indicate how much the event or situation happens to you. You are asked to respond by circling **one** number from 1 (Not at All) to 7 (To a Very Great Extent) that you feel best fits your experience. Please note that the zero value (0) is used only if the situation described does not apply to you. When answering these questions, we would like you to think about the organisation from which you first found out about this study. For easy reference, the name of that organisation is:

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**PEOPLE:** refers to those individuals who work directly with you or your child. These **may include** psychologists, therapists, social workers, doctors, teachers, etc.

IN THE PAST YEAR, TO WHAT EXTENT DO THE PEOPLE WHO WORK WITH YOUR CHILD...	Indicate <u>how much</u> this event or situation happens to you.							
	To a Very Great Extent	To a Great Extent	To a Fairly Great Extent	To a Moderate Extent	To a Small Extent	To a Very Small Extent	Not at All	Not Applicable
1. ...help you to feel competent a parent?	7	6	5	4	3	2	1	0
2...provide you with written information about what your child is doing in <u>therapy</u> ?	7	6	5	4	3	2	1	0
3...provide a caring atmosphere <u>rather</u> than just give you information?	7	6	5	4	3	2	1	0
4. ...let you choose when to receive information and the type of information you want	7	6	5	4	3	2	1	0
5. ...look at the needs of your “whole” child (e.g., at mental, emotional, and social needs) instead of just at physical needs	7	6	5	4	3	2	1	0
6. ...make sure that at least one team member is someone who works with you and your family over a long period of time	7	6	5	4	3	2	1	0
7. ...fully explain treatment choices to you?	7	6	5	4	3	2	1	0
8. ...provide opportunities for you to make decisions about treatment?	7	6	5	4	3	2	1	0

IN THE PAST YEAR, TO WHAT EXTENT DO THE PEOPLE WHO WORK WITH YOUR CHILD...	Indicate <u>how much</u> this event or situation happens to you.							
	To a Very Great Extent	To a Great Extent	To a Fairly Great Extent	To a Moderate Extent	To a Small Extent	To a Very Small Extent	Not At All	Not Applicable
9. ...provide enough time to talk so you don't feel rushed?	7	6	5	4	3	2	1	0
10. ...plan together so they are all working in the same direction?	7	6	5	4	3	2	1	0
11. ...treat you as an <u>equal</u> rather than just as the parent of a patient (e.g., by not referring to you as "Mom" or "Dad")	7	6	5	4	3	2	1	0
12. ...give you information about your child that is consistent from person to person?	7	6	5	4	3	2	1	0
13. ...treat you as an individual rather than as a "typical" parent of a child with a disability?	7	6	5	4	3	2	1	0
14. ...provide you with written information about your child's progress?	7	6	5	4	3	2	1	0
15. ...tell you about the results from assessments?	7	6	5	4	3	2	1	0

**ORGANISATION:** refers to all staff from the health care organisation, whether involved directly with your child or not. In addition to health care people they **may include** support staff such as office staff, housekeepers, administrative personnel, etc.

IN THE PAST YEAR, TO WHAT EXTENT DO THE PEOPLE WHO WORK WITH YOUR CHILD...	Indicate <u>how much</u> this event or situation happens to you.							
	To a Very Great Extent	To a Great Extent	To a Fairly Great Extent	To a Moderate Extent	To a Small Extent	To a Very Small Extent	Not At All	Not Applicable
16. ...give you information about the types of services offered at the organisation or in your community?	7	6	5	4	3	2	1	0
17. ...have information available about your child's disability (e.g., its causes, how it progresses, future outlook)?	7	6	5	4	3	2	1	0
18. ...provide opportunities for the entire family to obtain information?	7	6	5	4	3	2	1	0
19. ...have information available to you in various forms, such as a booklet, kit, video, etc.?	7	6	5	4	3	2	1	0
20. ...provide advice on how to get information or to contact other parents (e.g., organisation's parent resource library)?	7	6	5	4	3	2	1	0

## Appendix M: Demographic questionnaire.

**Title of study:** 'An exploration of the experience of caring for children with non-malignant life-limiting conditions including, accessing services and decision-making surrounding the child's care'.

**Sex** (Please tick)

Male

Female

**What is your date of birth?**

Day    Month    Year

**What is your current marital status?** (Please tick one box only)

Single (never married)

Married

Separated

Divorced

Widowed

Other (please specify) -----

**How many children do you have?** (Under eighteen years) .....

**5 What is the highest level of education/ training which you have completed to date?**

(Please tick one box only)

No formal education

Primary education

Secondary/ grammar education to GCSE/CSE/O Level

Secondary/ grammar education to A level

Technical or Vocational courses

Bachelor Degree or National Diploma

Professional qualification

Postgraduate courses

**How would you describe your present principal status?**

- Working for payment or profit
- Seeking employment
- Unemployed
- Student
- Looking after home/family
- Full time carer
- Retired from employment
- Unable to work due to permanent illness or disability
- Other (please specify) -----

**What is your ethnic or cultural background?**

- White
- Black or Black Irish
- Asian or Asian Irish

**Do you speak a language other than English or Irish at home?**

- Yes
- No

**Do you live in an urban or rural area?**

- Urban
- Rural area

**What type of accommodation does your household occupy?**

- House
- Flat or apartment
- Bedsit
- Caravan/ Mobile or temporary dwelling
- Other (please specify) -----

**Does your household own or rent your accommodation?**

- Own outright
- Own with mortgage
- Rent

**What age is your child that is accessing palliative care services today?**

- year(s)    months

**Is your child in receipt of respite care?**

- Yes
- No

**If yes how many hours per week?**  hours

**Have you encountered any difficulties in relation to accessing respite care?**

- Yes
- No

If yes, please provide a brief description

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**Where is the respite provided?**

- In the family home
- Outside the family home

**How many miles are you away from the hospital that your child attends?**

- miles

**How do you and your child usually travel to hospital appointments?**

- On foot
- Private car
- Taxi
- Bus
- Train

Other (please specify) -----

**How many cars are owned or are available for use by one or more members of your household?**

Car(s)

**Do any of your children other than (name of child) have healthcare concerns?**

Yes

No

If yes, please provide a brief description

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-----

**Do you provide regular unpaid personal help for a friend or family member with a long-term illness, health problem or disability?**

Yes

No

**If yes how many hours per week?**

hours

## Appendix N: Study summaries

Author, Year and Country	Aim/objective	Study design	Sample	Data collection techniques	Key findings	Comments
1. Bailey-Pearce et al. (2017) United Kingdom	Explore fathers' experiences of their child's life limiting illness. In addition explore complex attachment strategies utilised by fathers throughout the course of their child's illness.	Qualitative.	7 fathers of children diagnosed with a life-limiting condition for at least 1 year.	In-depth interviews. Fathers' attachment strategies were assessed using the Adult Attachment Interview.	<ul style="list-style-type: none"> <li>• The dominant themes were found to be 'experience of the diagnosis', 'living with the illness', 'struggling with emotions' and 'relationship with staff'.</li> <li>• Within each theme, there were differences which related to the father's attachment strategies.</li> <li>• This was particularly evident in parts of their narratives recounting critical moments of threat and anxiety in the course of discovering and adjusting to their child's illness.</li> <li>• Importantly, the findings also suggested that the experience for the fathers stressed, and in some cases disrupted, their attachment coping strategies.</li> <li>• All fathers told stories of trying to get it right for their children and family.</li> </ul>	<p>This study provides a vital insight into fathers' experiences of their child's life limiting illness. The number of fathers who participated was small. Additionally the range of conditions included in the study was not made clear. It was, however apparent from the narratives that many of the children had non-malignant life-limiting conditions.</p>

<p>2. Bally et al. (2018) Canada</p>	<p>To conduct a metasynthesis of qualitative research exploring parents' psychosocial experiences during complex and traumatic life transitions related to caring for a child with a life-limiting (LLI) or life-threatening illness (LTI).</p>	<p>Metasynthesis</p>	<p>A systematic search of qualitative research was conducted by an experienced librarian to identify and retrieve studies from 10 databases.</p>	<p>Of the 3,515 studies screened, 23 were included.</p>	<ul style="list-style-type: none"> <li>• A synthesis of the findings demonstrated that parents experience profound and pervasive uncertainty, leading to their own illness experience being described as a dual reality in which fighting for survival and recognizing the threat of their child's death were daily challenges.</li> <li>• Three key processes emerged: the devastation of living with uncertainty, the emergence of hope, and moving forward.</li> </ul>	<p>The majority of the studies included in this metasynthesis focused on childhood cancer (n = 14), maternal (n = 20), and North American (n = 13) samples, so the findings may not be transferable to populations and settings that differ.</p>
<p>3. Brennan, Hugh-Jones and Aldridge (2012) United Kingdom</p>	<p>Explore the experiences of siblings of children with life-limiting conditions and how they cope with those experiences.</p>	<p>Qualitatively driven, longitudinal, mixed methods approach.</p>	<p>31 healthy children (aged between 5 and 16 years) from 21 families.</p>	<p>Participants were visited once a year over 3 years. At the first visit, a brief demographic questionnaire and standardised measures were completed. At the second and third visits, qualitative data were collected from the participant, and</p>	<ul style="list-style-type: none"> <li>• Emotional functioning and perceptions of self-worth were normative on standardised measures.</li> <li>• Qualitative data indicated distinct psychosocial strategies that appeared to underpin functioning, positioning themselves as adults within the family, adopting a role of 'social glue' in key relationships and thereby</li> </ul>	<p>The authors highlighted a number of limitations. In particular, the included sample is unlikely to be representative of all siblings; it may be that only those families who were coping well agreed to participate.</p>

				standardised measures were repeated.	<p>diminishing their own needs, and compartmentalising home and school life.</p> <ul style="list-style-type: none"> <li>• Some strategies appeared adaptive in the short term but may be limiting in the long term.</li> </ul>	
4. Cadell, Kennedy and Hemsworth (2012) Canada	Explore posttraumatic growth in parent caregivers of a child with a life-limiting illness.	Mixed method in two phases.	In total, 273 completed questionnaires were returned from parent caregivers. 47 interviews (23 individual and 12 couple interviews) were conducted with parent caregivers who had completed the questionnaire.	The questionnaire in the first phase was comprised of eight instruments overall. The second phase consisted of semi-structured interviews.	<ul style="list-style-type: none"> <li>• Two overarching themes were prominent in both the quantitative and qualitative data.</li> <li>• The first describes stress related to financial burden associated with caregiving.</li> <li>• The second theme concerns the posttraumatic growth experienced by the parent caregivers.</li> </ul>	Included parent caregivers who were currently caring for a child with a life-limiting illness aged 19 or younger.
5. Caicedo (2014) United States	Examine physical health, mental health, family functioning and care burden of parent caregivers for medically complex, medical technology–dependent children.	Longitudinal study.	A total of 84 parents or guardians caring for a medically complex and/or technology-dependent child were recruited.	Parent Physical, Mental Health, and Family Functioning were measured using the Pediatric Quality of Life (PedsQL) Family Impact Module.	<ul style="list-style-type: none"> <li>• Physically, parents were tired.</li> <li>• Mentally, they were frustrated, anxious, and angry.</li> <li>• Socially, they felt isolated and that people did not understand their family situation.</li> <li>• They found it hard to talk with others including physicians and nurses.</li> <li>• Average weekly hours of direct care was 33.0 (SD = 30.4) hours.</li> </ul>	Following an initial face-to-face meeting data were collected once a month for 5 months. The present study used a convenience sample and may have resulted in self-selection.

<p>6. Carnevale et al. (2006) Canada</p>	<p>Explore the moral dimension of family experience through detailed accounts of life with a child who requires assisted ventilation at home.</p>	<p>Qualitative.</p>	<p>12 families (38 family members) of children who are supported by a ventilator at home.</p>	<p>Semi-structured interviews.</p>	<ul style="list-style-type: none"> <li>• Data analysis identified 6 principal themes: (1) confronting parental responsibility; (2) seeking normality; (3) conflicting social values; (4) living in isolation; (5) the voice of the child; and (6) questioning the moral order.</li> <li>• An overarching phenomenon that best characterizes these families' overall experiences was identified: daily living with distress and enrichment.</li> </ul>	<p>Children in the study population fell into 4 diagnostic groups: (1) abnormal ventilatory control (eg, central hypoventilation syndrome), (2) neuromuscular disorders, (3) spina bifida, and (4) craniofacial or airway abnormalities resulting in upper airway obstruction.</p>
<p>7. Carter, Edwards and Hunt (2015) United Kingdom</p>	<p>Explore key aspects of the work of the family support workers in caring and supporting families of life-limited children from the families' perspectives.</p>	<p>Mixed method.</p>	<p>55 families (12 bereaved) and 39 children aged 2–18 years.</p>	<p>Survey, interviews and ethnographic observation.</p>	<p>Thematic analysis revealed how the family support workers became a presence in families' lives in three main ways:</p> <ul style="list-style-type: none"> <li>• Encompassing and embracing families through supporting needs and promoting resilience.</li> <li>• Befriending and bonding through developing knowledge, trusting relationships and a sense of closeness.</li> <li>• Accompanying and enduring by 'being with' families in different settings, situations and</li> </ul>	<p>Included children with a wide range of life-limiting conditions including degenerative neurological conditions, muscular disease, metabolic and cardiac conditions. Also included children with malignant conditions. The exact breakdown of the numbers is not provided.</p>

					crises and by enduring alongside the families.	
8. Collins et al. (2016) Australia	Provide an in-depth exploration of the prevalent lived experiences of parents who are currently providing care for a child with a life-limiting condition.	Cross-sectional, prospective qualitative design.	14 parents (12 mothers and 2 fathers) who identified as the 'primary caregiver' for one or more children diagnosed with a life-limiting condition.	Semi-structured face-to-face interviews.	<ul style="list-style-type: none"> <li>• Four key themes represented the prevalent experiences of parents: (1) trapped inside the house, (2) the protector, (3) living with the shadow and (4) travelling a different pathway.</li> <li>• They describe parents' physical and social isolation, exclusion from the workforce, pervasive grief and associated impacts to their health and well-being.</li> <li>• Limited professional and diminished social supports resulted in full ownership of care responsibility.</li> <li>• Yet, parents embraced their role as 'protector', reporting acquired meaning and purpose.</li> </ul>	This study provides valuable information regarding the lived experiences of parents who are currently providing care for a child with a life-limiting condition. The findings revealed pertinent issues relating to access in particular the limited professional support.
9. Courtney et al. (2018) Ireland	Explore mothers' perspectives of the experiences and impact on themselves and their family when their child has a life-	Qualitative	12 mothers of children with severe neurodevelopmental delay.	Semi-structured interviews. 10 mothers requested a home interview; 2 requested a	<ul style="list-style-type: none"> <li>• Four themes were identified in relation to mothers' perspectives of their experiences and their impact on themselves/their</li> </ul>	The children with life-limiting neurodevelopmental disability ranged from 1 to 6 years. One child's diagnosis was unknown; five children

	limiting neurodevelopmental disability.			phone interview.	<p>family when their child has a life-limiting neurodevelopmental disability.</p> <ul style="list-style-type: none"> <li>• ‘Starting out’ captures mothers’ experiences of the birth of their child and the aftermath.</li> <li>• ‘Keeping the Show on the Road’ describes the strategies employed to manage life day to day and the resources used to do this.</li> <li>• ‘Shouldering the Burden’ describes the range of physical, psychological and social consequences of the situation for mothers and the family.</li> <li>• The ‘Bigger Picture’ relates to the world outside the family and how this too must be navigated and managed.</li> </ul>	<p>had an organic central nervous system disorder, whereas the remainder had a chromosomal abnormality. Although the specific diagnosis was sometimes unclear, it was reported that for all children, the problems were present from birth or thereafter. All received a variety of services including paediatric, intellectual disability and palliative care. The children experienced multiple health problems.</p>
10. Cote-Arsenault and Denney-Koelsch (2011) United States	Clarify the experiences and needs of families in order to design responsive perinatal palliative care services, and to establish the feasibility and acceptability of	Qualitative descriptive study.	2 women and 3 couples during pregnancy or just after birth. Total of 8 parents.	Interviews, using open ended questions.	<ul style="list-style-type: none"> <li>• Two dimensions and six themes emerged.</li> <li>• In the dimension of Personal Pregnancy Experience, ‘Grieving Multiple Losses’ elucidates that parents grieve the loss of their normal</li> </ul>	<p>This study was included in the review as one mother had a live birth and the infant’s condition was documented as ‘better than expected’. The infant had a life-limiting</p>

conducting intensive interviews of pregnant women and their partners during their pregnancy with a lethal fetal diagnosis.

- pregnancy, healthy baby and future parenting.
- 'Arrested Parenting' describes their sudden interruption in the normal process of becoming a parent.
  - The theme 'My Baby is a Person' reflects parents unanimous desire to honor and legitimize the humanity of their unborn baby.
  - In the dimension of Interactions of Others, three themes were found.
  - 'Fragmented Health Care' describes parent's disjointed and distant encounters with multiple providers.
  - 'Disconnected Families and Friends' describes the lack of understanding of what the families were experiencing.
  - 'Utterly alone,' which crosses both dimensions, expresses how the parents' sense of social isolation adds to their personal

condition. Valuable information regarding service provision is also presented in the study findings.

					sense of loss and loneliness.	
11. Davies (2005) Wales	Explore the loss of a child from parents' perspectives.	Qualitative.	10 mothers whose child had died from a life-limiting condition in the preceding three years.	One-to-one interview.	<ul style="list-style-type: none"> <li>• Four themes emerged: time to be with their dying child; space and privacy to be with their dying child; time to be with their child's body after death; and space and privacy to be with their child's body after death.</li> <li>• Memories of these events continued to affect them, giving further support to new theoretical understandings of parental grief which suggest that parents maintain continuing bonds with their dead child by preserving memories and recollections of their life and death.</li> </ul>	The research participants comprised of five mothers with access to a children's hospice and five mothers who lived in an area which did not have a children's hospice. The children had died from malignant and non-malignant life-limiting conditions.
12. Davies, Davis and Sibert (2003) Wales	Explore parents' experiences of care by paediatricians in the time leading up to and including diagnostic disclosure of a life-limiting condition in their child.	Qualitative.	30 families whose child was diagnosed with a life-limiting condition.	In-depth interviews.	<ul style="list-style-type: none"> <li>• Parents' stories centred upon their need to have their concerns and needs as parents validated by paediatricians.</li> <li>• Demonstrable qualitative differences were shown to exist between the sensitive and insensitive clinical</li> </ul>	Parents' stories provide evidence of the impact of practices by paediatricians upon them during what is a series of stressful life changing events.

					practices of paediatricians.	
13. Davies et al. (2004) United States	Enhance understanding of the experiences of fathers whose children were diagnosed with a life-limiting illness.	Retrospective grounded theory study.	8 fathers whose child had died of a life-limiting illness.	In-depth, unstructured interviews.	<ul style="list-style-type: none"> <li>• Every aspect of fathers' lives was affected by their experiences, which were described in metaphoric terms as living in a dragon's shadow.</li> <li>• Fathers dealt with life in the dragon's shadow by battling the dragon (the illness) –the core social process.</li> <li>• Battling was a conscious, active, continuous process that required strength, willpower, and work.</li> <li>• Battling occurred within the context of fathers' experiences with fathering and fatherhood and was characterised by 3 aspects: battling with uncertainty, battling with responsibility, and battling with everyday disruption.</li> <li>• Fathers were assisted by supportive work environments and by supportive relationships with health care providers.</li> </ul>	This study provides an invaluable insight into father's perspectives of living with and losing a child to a life-limiting illness. The number of fathers who participated was small with the majority having a child with a malignancy. The findings reveal issues surrounding access in particular relationships with healthcare providers.

					<ul style="list-style-type: none"> <li>Unsatisfactory relationships with medical personnel compounded fathers' battling with life in the dragon's shadow.</li> </ul>	
14. Dybwik et al. (2011) Norway	Explore the experiences of families giving advanced care to family members dependent on home mechanical ventilation.	Qualitative grounded theory.	A total of 15 family members with 11 ventilator-dependent individuals (3 children and 8 adults). Although it was not explicitly stated the 3 children appeared to have a life-limiting diagnosis.	In-depth interviews.	<ul style="list-style-type: none"> <li>The core category, 'fighting the system,' became the central theme as family members were asked to describe their experiences.</li> <li>Three subcategories were identified. These included 'lack of competence and continuity', 'being indispensable' and 'worth fighting for'.</li> <li>This study revealed no major differences between the families caring for a child or an adult.</li> </ul>	This study provides a valuable insight into the experiences of families giving advanced care to family members. However the, majority of the participants were caring for adults. Although the findings revealed no major differences between the families caring for a child or an adult the findings cannot be generalised to families giving advanced care to children. Pertinent issues surrounding access to services were raised.
15. Eaton (2008) United Kingdom	To describe the experiences of families, whose children have life-limiting and life-threatening conditions and who have complex healthcare	Qualitative.	11 families of children (aged between 3 and 16 years) receiving respite care in one of two environments, hospice or home.	Semi-structured interviews.	<ul style="list-style-type: none"> <li>The areas of concern identified as significant to all the families were referral to respite service, service organisation, communication, relinquishing</li> </ul>	The findings revealed significant issues regarding service provision. Conditions included epilepsy, cerebral palsy

	needs, of receiving respite care at home or in a hospice.				control to respite carers and satisfaction with service.	and complex special needs.
16. Gaab, Owens and MacLead (2013) New Zealand	Explore how primary caregivers communicate with their children about their impending mortality.	Qualitative.	19 primary caregivers of children receiving palliative care or who had received palliative care to their death. 10 of the children had cancer, 5 had heart conditions, 3 had muscular dystrophy and the 1 child's condition was not disclosed.	Semi-structured in-depth interviews.	<ul style="list-style-type: none"> <li>Caregivers face a number of common challenges and concerns during and after caring for a child receiving pediatric palliative care which is related to much more than their children's impending deaths.</li> <li>Primary caregivers spoke about family support, regrets/feeling of blame, disability discrimination, parental roles, internet research, helpful organisations, and research advice.</li> </ul>	Although this research set out to explore how primary caregivers communicate with their children about their impending mortality, many other experiences arose from the interviews. The cohort that caregivers cared for did not represent the wide range of life-limiting conditions.
17. Gilmer et al. (2013) United States	Describe parental perceptions of the care of hospitalized, terminally ill children in the areas of (1) clinical management, (2) interdisciplinary support, and (3) pain and symptom management.	Mixed method.	Phase 1 Medical records of all children who died during one year were examined (N= 105). Phase 2 Bereaved parents (N= 15).	Phase 1 Medical records. Phase 2 Modified FAMCARE survey and semi-structured telephone interviews.	<ul style="list-style-type: none"> <li>Data provided evidence of areas which lack satisfaction with elements of end-of-life care of hospitalized children.</li> <li>Parental satisfaction with their child's care during end of life indicates need for improvement in pain management, communication, and parental</li> </ul>	Phase 2 included 14 mothers and 1 father. Admitting diagnoses included: cardiac diagnoses, congenital defects, neonatal-specific diagnoses, infectious diseases and oncology diagnoses.

					involvement in decision making.	
18. Goldstein et al. (2013) Switzerland	Explore fathers experiences of their child's chronic illness.	Literature review.	Using PubMed, a search was undertaken using the following MeSH terms: Adolescent, Father, Asthma, Cystic Fibrosis, Diabetes, Juvenile Rheumatoid Arthritis and Neoplastic Disease. The search was performed on the 1 <sup>st</sup> of June 2010, and returned 152 results.	33 articles were selected as appropriate and analysed.	<ul style="list-style-type: none"> <li>• Three time-periods describe the obstacles fathers tackle when parenting children with chronic disease: a) diagnosis and short-term characterized by distress, isolation and uncertainty; b) the mastery period, characterized by the struggle to establish routine and c) the long-term, characterized by relationship and personality change, worries and bereavement.</li> </ul>	This review had some limitations. In particular, the search was limited to one database (PubMed).
19. Gupta and Prescott (2012) United Kingdom	Determine the impact of services for newly referred children with non-malignant life-limiting and life-threatening conditions and their families, in terms of parental stress and psychological wellbeing.	Quantitative.	Data was collected on all new referrals to the children's palliative care partnership. This included a total of 59 families.	Within a three year period demographic data was collected on all new referrals to the children's palliative care partnership. At the point of referral, all parents/guardians (one per family) were invited to complete the Parenting Stress Index and the	<ul style="list-style-type: none"> <li>• Statistical analysis indicated that there was no significant change at 12 month follow-up.</li> <li>• The lack of deterioration in levels of parental stress and psychological wellbeing is viewed positively within this context.</li> <li>• It is hypothesised that multi-agency and partnership working was a significant contributory factor in not increasing levels of parental stress</li> </ul>	The findings demonstrate the impact services can have on the families' experiences.

				General Health Questionnaire. The measures were repeated 12 months later.	and psychological wellbeing through social support.	
20. Heaton et al. (2005) United Kingdom	Explore families' experiences of caring for a technology-dependent child from a temporal perspective.	Qualitative.	36 families (75 family members) with technology dependent children who used one or more devices on a daily basis.	Interviews.	<ul style="list-style-type: none"> <li>The authors found that the rhythms and routines of care varied across the sample, depending on the type and number of devices used, the individual child's needs, and who provided technical care during the day and/or at night at home and in other settings.</li> <li>While the children's health and quality of life benefited from the technology, the time demands of the care routines and lack of compatibility with other social and institutional timeframes had some negative implications for the children and their families.</li> <li>The need to use and oversee the use of some medical technologies at night also meant that many parents suffered</li> </ul>	Devices included feeding pumps, suction machines, dialysis machines and ventilators. It was unclear as to how many children had life-limiting conditions.

					regular disruption to their sleep.	
21. Hobson and Noyes (2011) Wales	Describe the experiences of fathers who cared for their children with complex health and nursing care needs.	Qualitative.	8 fathers of children with complex health and nursing care needs (7 were biological fathers and 1 was a stepfather).	Face to face interviews using a broad topic guide.	<ul style="list-style-type: none"> <li>Fathers enjoyed their caring role and found it rewarding and at times stressful.</li> <li>They instituted structured regimes, which focused on the father/child/family.</li> <li>Performing intimate care posed specific challenges for which there is no guidance.</li> <li>Children's community nursing was highly valued.</li> <li>Fathers generally rejected the need for specific father-focused services.</li> <li>Fathers reported positive relationships with their children and partners.</li> </ul>	Four fathers were full-time carers and four were in full-time employment but heavily involved in their child's complex nursing care. It was unclear as to how many children had life-limiting conditions.
22. Hsiao, Evan and Zeltzer (2008) United States	Identify the aspects of physician communication that children with life-limiting illnesses and their parents perceived to be facilitative or obstructive in pediatric palliative care.	Qualitative.	20 parent and child pairs of pediatric oncology and cardiology patients (mean age 14.25 years, range 9-21 years) with a poor prognosis (physician reported likely <20% chance of survival beyond 3 years) from two children's	Semi-structured interviews.	<ul style="list-style-type: none"> <li>Both children and parents identified five domains of physician communication deemed to be highly salient and influential in quality of care.</li> <li>These included relationship building, demonstration of effort and competence, information exchange, availability, and</li> </ul>	This study is part of a larger mixed-methods study using both qualitative and quantitative research methodology to assess communication in pediatric palliative care. It may be difficult to generalise the findings from this study to all children with

			hospitals and one pediatric hospice.		<p>appropriate level of child and parent involvement.</p> <ul style="list-style-type: none"> <li>• Parents identified coordination of care as another important communication domain.</li> <li>• The characteristics of physicians that were deemed most harmful to satisfying communication included having a disrespectful or arrogant attitude, not establishing a relationship with the family, breaking bad news in an insensitive manner, withholding information from parents and losing their trust, and changing a treatment course without preparing the patient and family.</li> </ul>	life-limiting conditions as the focus was on oncology and cardiology patients.
23. Knapp et al. (2010) United States	To determine how families are impacted by their children's illnesses.	Quantitative.	85 parents whose children had life-limiting illnesses and were enrolled in Florida's publicly funded pediatric palliative care program.	Cross-sectional telephone survey.	<ul style="list-style-type: none"> <li>• For 11 of the 15 Impact on Family (IOF) questions, the results showed that half or more of parents agreed with items on the IOF.</li> <li>• For example, 65% or more of parents agree that their child's illness results in</li> </ul>	The majority of parents who responded to the survey were white non-Hispanic (56%), married (54%), had graduated from college (33%), and live in a two-parent household (60%).

					<p>parental fatigue (71%), giving up things (71%), having to change plans at the last minute (76%), difficulties finding a reliable caregiver (77%), and that living with an ill child can sometimes feel like a rollercoaster ride (90%).</p> <ul style="list-style-type: none"> <li>• Multivariate analysis showed that having depressive symptoms was associated with greater impact on the family.</li> </ul>	
24. Lahtinen and Joronen (2014) Finland	To describe parents' experiences of caring for a ventilator-dependent child at home.	Literature review.	The data consisted of articles searched systematically from Medic, Arto, Linda, CINAHL, Medline and PsycINFO databases and manually from years 1998-2012.	18 articles were selected.	<ul style="list-style-type: none"> <li>• Parents' experience of caring for a ventilator-dependent child at home was viewed as a constant battle of survival.</li> <li>• The constant battle of survival consisted of struggling with life management challenges and maintaining the balance in family functioning.</li> <li>• The home became like hospital because of the technology and the presence of the health care staff.</li> <li>• Parenting a ventilator child also included the</li> </ul>	It was unclear as to how many children had life-limiting conditions.

					dimension of nursing.	
25. Lazzarin et al. (2018) Italy	Investigated home-based palliative care for young children and how long it took parents to meet their needs.	Quantitative.	33 Parents (30 mothers and 3 fathers) caring for children with life-limiting conditions.	Telephone interviews using ad hoc structured interview questions that concerned the procedures and medical appliances used at home and the amount of time, in minutes, spent on daily care.	<ul style="list-style-type: none"> <li>The children needed an average of five different life-supporting medical appliances.</li> <li>The time taken to provide their care increased significantly with each additional appliance (<math>p = 0.016</math>).</li> <li>Their most time-consuming daily needs were feeding (174 minutes) and support when they woke up at night (67 minutes).</li> <li>The average daily time that parents spent taking care of their child amounted to eight hours and 54 minutes.</li> </ul>	This study provides valuable information regarding the amount of time parents spend managing their child's basic healthcare needs, such as personal hygiene and feeding, administering drugs and managing symptoms. This was without taking into account the other normal activities that the parents engaged in with their children, such as playing, entertainment and schooling.
26. Malcolm et al. (2013) United Kingdom	Understand the family experience of supporting a child with the rare degenerative life-limiting conditions (LLCs) of MPS and Batten disease.	Qualitative.	8 siblings of children with MPS ( $n = 7$ ) and Batten Disease ( $n = 1$ ).	Semi-structured interviews.	<ul style="list-style-type: none"> <li>Siblings are clearly impacted emotionally, pragmatically and relationally by the ill health of another child in the family.</li> <li>The data indicate four key themes which demonstrate impacts on siblings: perceptions of the condition and its symptoms, impact on daily life, emotional</li> </ul>	This paper presents data from a UK-wide study that sought to understand the family experience of supporting a child with the rare degenerative LLCs of MPS and Batten disease. The aim of this paper is to report sibling experiences related to these

					<p>consequences and ways of coping.</p> <ul style="list-style-type: none"> <li>• Siblings often had considerable knowledge of the condition and took on important roles in symptom management.</li> <li>• However, these experiences were in the context of managing relationships within the family (often protecting parents from an awareness of how much they knew) and relationships at school (including distraction from learning and being bullied by peers).</li> </ul>	<p>rare degenerative and progressive conditions.</p>
<p>27. Meert et al. (2008) United States</p>	<p>Describe parents' perceptions of their conversations with physicians regarding their child's terminal illness and death in the pediatric intensive care unit (PICU).</p>	<p>A secondary analysis of a qualitative interview study.</p>	<p>56 parents of 48 children who died in the PICU 3–12 months before the study.</p>	<p>Semi-structured telephone interviews.</p>	<ul style="list-style-type: none"> <li>• The most common communication issue identified by parents was the physicians' availability and attentiveness to their informational needs.</li> <li>• Other communication issues included honesty and comprehensiveness of information, affect with which information was provided, withholding of information, provision of false</li> </ul>	<p>Limitations of this study include the large number of parents who could not be contacted and the predominance of mothers among participants. This study includes only bereaved parents; it is plausible that the views of parents who are not bereaved may be different.</p>

					hope, complexity of vocabulary, pace of providing information, contradictory information, and physicians' body language.	
28. Menezes (2010) United Kingdom	Understand the perceptions and experiences of the child with a life-limiting condition, and to frame that understanding with family insights.	Qualitative.	11 children and young people affected by life-limiting conditions. Parents and siblings were also vital contributors (39 participants from 10 families).	Topic-guided interviews and participant observation. Participants (especially the children and their siblings) were invited to use their own artwork or photographs (either from the family collection or specifically taken for the research) to help explain their day-to-day experiences.	<ul style="list-style-type: none"> <li>This study identified several 'moments of realization', representing times in the child's life when participants recognized the real threat to the child's life.</li> <li>These included: questions of inheritance, diagnosis and prognosis, acute loss of abilities, slow deterioration and life-threatening surgery.</li> </ul>	The author used case study to provide the overarching approach. It is not clear to what extent the participants might represent other life-limited children and their families.
29. Moola (2012) Canada	Explore the experiences of parents of children with cystic fibrosis and parents of children with congenital heart disease and gain insight regarding the similarities and differences in the parenting experiences.	Qualitative.	29 parents from separate families participated. This included 10 fathers and 19 mothers.	Semi-structured interviews.	<p>Three main themes are reported in this study.</p> <ul style="list-style-type: none"> <li>First, although a sense of stress was ever present for all parents, parents of children with congenital heart disease and cystic fibrosis narrated stress in different ways.</li> <li>Second, the temporal losses associated with both the time-consuming</li> </ul>	16 parents of children with cystic fibrosis and 13 parents of children with congenital heart disease participated.

					<p>nature of treatment and the lifelimiting nature of the disease were specific sources of stress for parents of children with cystic fibrosis only.</p> <ul style="list-style-type: none"> <li>• Finally, parents of both children with cystic fibrosis and children with congenital heart disease dealt with the chronic stress of caregiving by coming to terms with their child's illness and learning to put things into perspective</li> </ul>	
30. Nicholas et al. (2016) Canada	<p>Explore the experiences of fathers of children with a life-limiting illness. Explore their preferences regarding support provision.</p>	<p>Qualitative grounded theory.</p>	<p>18 fathers of children with life-limiting illness (6 fathers had experienced the death of their child).</p>	<p>In-depth, semi structured interviews either one-to-one or focus groups. Two focus groups were conducted: one for bereaved fathers and the other for non-bereaved fathers.</p>	<ul style="list-style-type: none"> <li>• The overarching themes were stresses, means of coping, and perceived needs for support.</li> <li>• Generally, fathers struggled relative to discursive and internalised notions of fathers as providers and protectors for their children, combined with an inability to ease their child's vulnerability to life-limiting illness.</li> <li>• Fathers were engaged in the care of their child with life-limiting illness, but</li> </ul>	<p>This study provides an invaluable insight into father's perspectives of living with and losing a child to a life-limiting illness. The number of fathers who participated was significant. The primary diagnoses of the children varied— including cancer, cystic fibrosis, cerebral palsy, enzyme deficiency, and</p>

					<p>several felt marginalised by healthcare providers.</p> <ul style="list-style-type: none"> <li>• Some fathers recognised and valued their support network while others had few supports.</li> <li>• Some described personal growth and desired to help others.</li> </ul>	congenital heart defects.
31. Nicholl et al. (2013) Ireland	Identify the range of technology used by children with complex needs and identify issues of concern for parents.	Mixed method.	<p>Phase 1 9 nurses recruited from a voluntary organisation involved in providing home respite services to children up to 4 years of age with complex needs and associated neurodevelopmental disabilities.</p> <p>Phase 2 177 parents of children with complex needs.</p>	Data in phase one focused on the development of an appropriate questionnaire and were collected during a focus group interview with a purposive sample of expert nurses. The questionnaire was subsequently administered by telephone survey and personal contact with parents of children with complex needs.	<ul style="list-style-type: none"> <li>• Findings indicated that children with intellectual disabilities and associated complex needs require the use of high levels of technology to maintain their health and well-being.</li> <li>• On average, families identified that 22 pieces of equipment were used by their child.</li> </ul>	The study comprised of two phases, qualitative and quantitative. Both designs complement each other with Phase 2 dependent upon Phase 1. The authors outline that while the qualitative findings are being prepared for publication, the focus of this paper is the quantitative findings.

32. Parravicini et al. (2018) United States	To assess the perception of parents concerning the state of comfort maintained in their infants born with life-limiting conditions and treated by a standardised neonatal comfort care program.	Quantitative.	42 parents (26 mothers and 16 fathers) who elected comfort care for their newborns diagnosed with life-limiting conditions. The 42 parents represented 35 families.	Questionnaire with quantitative response options and open-ended questions.	<ul style="list-style-type: none"> <li>• Most parents reported that their child was treated with respect, in a caring, peaceful and non-invasive environment.</li> <li>• To the question 'Do you think that overall your baby received comfort' mode response was 'always'.</li> </ul>	Standardised comfort measures including family/infant bonding, warmth, nutrition/hydration and pain/discomfort management were provided to all infants.
33. Popoviciu et al. (2013) Romania	Explore parental challenges when children were diagnosed with life-limiting illnesses.	Qualitative.	30 parents (27 mothers and 3 fathers) whose children were referred to one hospice that has a paediatric team working in close collaboration with the oncology-hospital unit.	Interviews.	Interview analysis revealed four overarching themes: (a) financial burdens, (b) lack of information about the disease, (c) lack of social understanding/support and (d) guidance in giving children information about their illness.	Children with non-malignant and malignant life-limiting illnesses were included. While not without limitations (i.e. possible interviewer influence on parental perceptions/reports; small purposive sample), this research adds to our understanding of the unique challenges of parents in a context of poverty.
34. Rallison and Raffin-Bouchal (2013) Canada	Explore the experience of families caring for a child with a progressive neurodegenerative illness at home.	Qualitative.	6 families comprised of 27 family members participated in the study. A total of 6 children were observed.	In-depth interviews and observation.	<ul style="list-style-type: none"> <li>• The illness journey was revealed to be complex and unpredictable.</li> <li>• The three main discoveries were the families' experiences of holding predictability and</li> </ul>	Family members in the study included parents, siblings, the ill child, and other significant individuals that the family identified as their family.

					<p>unpredictability, the complexity and relentless care in the home, and the spiritual journey of the families.</p>	
<p>35. Rodriguez and King (2008) United Kingdom</p>	<p>Explore the lived experience of parenting a child with a life-limiting condition: a focus on the mental health realm.</p>	<p>Qualitative.</p>	<p>10 parents (all mothers) of children with life-limiting conditions.</p>	<p>Semi-structured interviews.</p>	<ul style="list-style-type: none"> <li>The essential meaning of the phenomenon 'the lived experience of parenting a child with a life-limiting condition' is understood as a full-time emotional struggle involving six continuous constituents: inner drive, feeling responsible, psychological effects, threatened self-image, social withdrawal, and a fear of reaching the bottom line.</li> </ul>	<p>The majority of children had non-malignant life-limiting conditions.</p>
<p>36. Smith, Cheater and Bekker (2015) United Kingdom</p>	<p>Investigate parents' experiences of living with a child with a long-term condition using systematic methods.</p>	<p>A rapid structured review of the literature.</p>	<p>Three data bases (MEDLINE, CINAHL, PSYCINFO) were searched 1999-2009.</p>	<p>34 studies were included in the review.</p>	<ul style="list-style-type: none"> <li>The impact of living with a child with a long-term condition related to dealing with immediate concerns following the child's diagnosis and responding to the challenges of integrating the child's needs into family life.</li> <li>Parents' perceived they are not always supported in their quest for information and</li> </ul>	<p>The children had varying diagnoses. These included chronic conditions such as diabetes and life-limiting conditions.</p>

					<p>forming effective relationships with health-care professionals can be stressful.</p> <ul style="list-style-type: none"> <li>• Although having ultimate responsibility for their child's health can be overwhelming, parents developed considerable expertise in managing their child's condition.</li> </ul>	
37. Somanadhan and Larkin (2016) Ireland	Explore and interpret parents' experiences of living with and caring for children, adolescents and young people with Mucopolysaccharidosis (MPS).	Qualitative.	8 parents of children with a range of MPS disorders aged 6 months to 22 years.	Parents were interviewed at three points over a 17 month period.	<ul style="list-style-type: none"> <li>• The main themes identified during data analysis were described as living with MPS, living with a genetic rare disease, the stigma of a rare condition, MPS as encompassing multiple diseases, unknown future, hospital vs. home, experience of waiting, a tough road ahead, and things in their day-to-day life with MPS.</li> <li>• They spoke of their child's Quality of Life (QoL), their healthy children's wellbeing, and some, the impact on their own physical and psychological wellbeing.</li> <li>• They also reflected on issues of</li> </ul>	MPS is one of a range of rare inherited metabolic disorders that come under Category 3 of life-limiting conditions, where there is no curative treatment available at present.

					stigmatisation and isolation in their experience of living with a child with a rare disorder.	
38. Steele and Davies (2006) United States	Enhance understanding of the experiences of families with a child who has a neurodegenerative life-threatening illness.	Qualitative.	Data were collected from eight families (29 family members).	Observations and interviews.	<ul style="list-style-type: none"> <li>• The impact on parents was pervasive and multidimensional.</li> <li>• Parents faced many emotional, physical, financial, and spiritual impacts on their lives.</li> <li>• Parents often needed help to alleviate the impact of their experience, but it was not always available in a useful manner.</li> <li>• Despite the negative aspects, caring for the child was never viewed by parents as a burden.</li> <li>• Fatigue was a particularly prevalent symptom that held the potential to impact on parents in multiple ways.</li> </ul>	The findings from this study reveal valuable insights regarding the experiences of families and their interaction with service providers.
39. Verberne et al. (2017) Netherlands	Gain insight into parental caregiving based on the lived experience of parents with a child with a life-limiting disease.	An interpretative qualitative study.	42 parents of 24 children with a malignant or non-malignant disease, receiving paediatric palliative care.	Single or repeated interviews were undertaken.	<ul style="list-style-type: none"> <li>• Based on their ambition to be a 'good parent', parents caring for a child with a life-limiting disease strived for three aims: controlled symptoms and controlled disease, a life worth living for</li> </ul>	9 of the children had a malignant disease and 15 had a non-malignant disease.

					<p>their ill child and family balance.</p> <ul style="list-style-type: none"> <li>• These aims resulted in four tasks that parents performed: providing basic and complex care, organising good quality care and treatment, making sound decisions while managing risks and organising a good family life.</li> </ul>	
40. Yogman and Garfield (2016) United States	Review new studies of the epidemiology of father involvement in the care and developmental of their children: the role of paediatricians.	Clinical report.	New studies of the epidemiology of father involvement, including nonresidential as well as residential fathers.	Not stated.	<ul style="list-style-type: none"> <li>• The effects of father involvement on child outcomes are discussed within each phase of a child's development.</li> <li>• Particular emphasis is placed on (1) fathers' involvement across childhood ages and</li> <li>• (2) the influence of fathers' physical and mental health on their children.</li> <li>• Implications and advice for all child health providers to encourage and support father involvement are outlined.</li> </ul>	This report explores fathers involvement with children with special healthcare needs. The authors concluded that the population of children with special health care needs has not been well studied and needs better research.
41. Zimmerman et al. (2016) Switzerland	To assess the end of life care perspectives of parents who had lost a child due to a cardiac, neurological or oncological	Quantitative.	135 parents.	The parental perspective was assessed with a newly developed and tested instrument that was	<ul style="list-style-type: none"> <li>• The findings show positive parental experiences of their child's end-of-life care and high perceived satisfaction with</li> </ul>	The four diagnostic groups were chosen as they represent the major diagnoses causing illness-

	condition, or during the neonatal period in the years 2011 or 2012.			structured according to six evidence-based quality domains.	<p>the care their child received.</p> <ul style="list-style-type: none"> <li>• Parents of a child with cancer rated their experiences in most of the six quality domains and reported the highest satisfaction with care.</li> <li>• The lowest scores were mainly reported by parents from the neurology group, with the exception of the shared decision making domain, where parents of neonates reported significantly less positive experiences.</li> </ul>	related death in children. This study was part of a larger research project concerned with paediatric end of life care in Switzerland
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Author, Year and Country	Aim/objective	Study design	Sample	Data collection techniques	Key findings	Comments
1. Bluebond-Langer et al. (2013) United States	To review the literature on preference for place of death for children and young people with life-limiting or life-threatening illnesses.	Systematic literature review.	The databases MEDLINE, CINAHL and EMBASE were searched from 2004-2012, as well as bibliography, key author and grey literature searches. Policy documents, empirical, theoretical and peer reviewed studies and conference abstracts were included.	9 studies were included from five countries.	<ul style="list-style-type: none"> <li>• Six studies reported a majority of parents (only one study interviewed adolescents) expressing preference for death at home.</li> <li>• Other studies differed significantly in their findings; one reporting 35.1% and another reporting 0% preferring death at home.</li> <li>• Some parents did not express a preference.</li> </ul>	Six of the studies included parents of children who died from cancer while being treated at tertiary centres that offered palliative care services. Therefore, these findings cannot be generalised to the entire population of children with life-limiting or life-threatening illnesses.
2. Brenner et al. (2015) Ireland	Explore parents' perspectives of the transition to home of a child with complex respiratory health care needs.	Qualitative.	Parents of children with a tracheostomy with or without other methods of respiratory assistance, who had transitioned home from a large Children's hospital in the last 5 years. 15 parents participated (12 mothers and 3 fathers), representing the transition to home of 15 children.	Telephone interview. Parents were asked three open-ended questions. Prompts were used where needed.	<ul style="list-style-type: none"> <li>• Four key themes which emerged from the interviews were 'stepping stones: negotiating the move to home', 'fighting and frustration', 'questioning competence' and 'coping into the future'.</li> <li>• Each of the parents expressed their anger and frustration at the length of time it took to have everything in place so they could take their child home.</li> <li>• Many of the parents</li> </ul>	This study explored the experiences of the transition to home of a child with complex healthcare needs. Although there was no reference to life-limiting it was apparent that the children had very complex medical needs. Additionally, many parents expressed concern regarding the future, suggesting that the future could

					<p>interviewed raised concern about the quality of care available in the community and in regional hospitals once they had been discharged from the tertiary care centre.</p> <ul style="list-style-type: none"> <li>• Parents spoke at length about their ongoing challenges and their hopes for the future. This included difficulty thinking beyond the present moment, concern for their child coping with their level of ability and the need for a compassionate health service to help parents cope.</li> </ul>	be very negative to contemplate.
3. Coad et al. (2015) United Kingdom	Explore the perceived met and unmet need of life-limited children, young people and families.	Qualitative Appreciative Inquiry.	59 adult individuals who were part of 51 families. This included 74% mothers; 13% fathers and remainder 13% were key family members (birth grandparents, foster grandparents and adoptive parents) who play a primary care role for the child or young person.	Semi-structured, in-depth interviews were conducted in the home setting or in a focus group setting, as preferred. In line with the Appreciative Inquiry approach, interviews included participatory arts-based methods to help participants to	<ul style="list-style-type: none"> <li>• Findings indicated that children and their families felt medical/nursing needs were well met but provision was needed for broader financial, social and emotional support alongside more responsive specialist therapies.</li> <li>• Children, young people and their families were living as normal lives as possible in abnormal circumstances.</li> </ul>	Two families that took part had two sons with life limiting/threatening conditions and two families were bereaved over two years prior to interview. The majority of the children/young people who participated had non-malignant life-limiting conditions.

			18 children and young people were also interviewed.	explore what was good about services (met needs); what could be better about services (unmet needs) and what the ideal for future services would look like.	<ul style="list-style-type: none"> <li>Family coping strategies included siblings, parents and carers such as grandparents and adult siblings, educating themselves and knowing all they need to know to help them to deal with what was happening.</li> </ul>	
4. Grinyer, Payne and Barbarachild (2010) United Kingdom	Elicit the views of service users on their experiences of respite care in a children's hospice.	Evaluation. The overall evaluation used a mixed method approach involving postal screening followed up by interviews, which are the focus of this paper.	24 service users (11 families) which included: parents, children and young people, siblings, guardians and family carers.	In-depth interviews.	<ul style="list-style-type: none"> <li>Lack of choice. There seemed to be little choice about when, how often, and for how long respite care was offered.</li> <li>Practical problems of access. The duration of the respite care was often very short-sometimes a single night-and the complicated preparations necessary were thought by some to be disproportionate.</li> <li>Powerless dependency. Parents were willing to endure difficulties and frustrations discussed above because they experienced the respite service as a 'lifeline'.</li> </ul>	This evaluation was confined to one children's hospice. Significant issues regarding access to services were highlighted. The findings demonstrate the tensions that parents in need of respite feel as a result of the power unconsciously exercised by staff. The children all had non-malignant life-limiting conditions.

<p>5. Jennings and Nicholl (2014) Ireland</p>	<p>Explore mothers' experiences of bereavement support following the death of their child from a life-limiting condition.</p>	<p>Qualitative.</p>	<p>10 mothers who had been bereaved in the previous 5 years.</p>	<p>Unstructured interviews.</p>	<ul style="list-style-type: none"> <li>• The findings from this study indicated that mothers relied on a combination of different sources of bereavement support.</li> <li>• The mothers relied on a combination of informal and formal bereavement support.</li> <li>• In addition to depending on others to provide support, the mothers described their ability to self-support.</li> <li>• The mothers supported themselves by taking steps to maintain the memory of the deceased child, which brought them comfort.</li> </ul>	<p>This study was limited by the use of a gatekeeper for recruitment, use of purposive sampling, and the fact that all of the mothers were English speaking. No claims are made as to the generalisability of the results.</p>
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<p>6. Kiernan et al. (2019) Ireland</p>	<p>Explore parents' experiences of service provision for their child with a life-limiting neurodevelopmental disability in an Irish context.</p>	<p>Mixed methods design.</p>	<p>63 parents of children under six years of age with a life-limiting neurodevelopmental disability completed a survey and interviews were conducted with 12 mothers who had completed the survey.</p>	<p>A postal survey was developed for this study. The survey also included The Measure of Processes of Care (MPOC-20). Semi-structured interviews.</p>	<ul style="list-style-type: none"> <li>• Results suggest parents' experiences are mixed, showing more negative than positive experiences.</li> <li>• Access to services was described as problematic, with obstacles including funding and geographical inequities.</li> <li>• Helpful aspects included staff attitudes and relationships and, to a lesser extent, coordinated care.</li> <li>• Unhelpful aspects included inconsistencies in the provision of services.</li> <li>• The findings suggest some implications for service provision, including the need for a more family-centred approach to care.</li> </ul>	<p>Most survey respondents were mothers (92%, n = 58). Given the dominance of mothers in the survey, it was decided to focus on this group for the interviews. It was decided to focus on children under the age of six, to ensure some homogeneity regarding the period of care the families had experienced to date.</p>
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<p>7. Kirk and Pritchard (2012) United Kingdom</p>	<p>Investigate parents' and young people's perceptions of hospice support and identify how support could be improved.</p>	<p>Mixed method.</p>	<p>A total of 108 postal questionnaires were returned from parents (71 parents and 37 bereaved parents) who had used the services of one hospice in the previous two years and interviews were conducted with 12 parents and 7 young people sampled from the postal questionnaire.</p>	<p>Postal questionnaires and in-depth qualitative interviews.</p>	<ul style="list-style-type: none"> <li>Families were highly satisfied with the support provided in terms of quality of care; interpersonal qualities of the staff; the individualized, family-focused approach; accessibility of support and involvement in decision making.</li> <li>Young people valued the opportunity to meet with other young people and take part in different activities.</li> <li>For parents the provision of a break from caring was the main way in which they were supported although they felt they wanted more of this form of support.</li> <li>A consistent theme in relation to support for young people and siblings was the need to develop facilities, activities and bereavement support specifically for teenagers/young people.</li> </ul>	<p>All of the children who used the hospice services had non-malignant life-limiting conditions. Information regarding the gender of parents who participated was not provided.</p>
<p>8. McGrath (2001) Australia</p>	<p>Obtain feedback on the need for a volunteer service for families coping</p>	<p>Quantitative.</p>	<p>107 families with a child diagnosed with a life-</p>	<p>A short (two page) questionnaire was developed to obtain</p>	<ul style="list-style-type: none"> <li>The questionnaire responses indicate that many families</li> </ul>	<p>Notably, over half of the questionnaires were returned from the Cystic</p>

	with a child with a life-limiting condition.		limiting condition.	feedback. Questions were asked about the perceived need for trained volunteers, what activities they could be used for, whether the need for them changes over time, obstacles to their use, and previous experience with volunteers.	<p>have only minimal or no support, and are coping with extraordinary physical, emotional and social demands from the child's condition and treatment.</p> <ul style="list-style-type: none"> <li>• The participants are very positive about the need for a trained volunteer and are clear about the activities that trained volunteers could provide assistance with.</li> <li>• These activities range from practical assistance such as baby-sitting and help with household chores and errands to emotional support.</li> <li>• Not all families would be comfortable using a volunteer, and some respondents did outline perceived obstacles to including a volunteer in family activities.</li> <li>• For others there were comments about why volunteers would be perceived as unproblematic.</li> </ul>	Fibrosis Association. Whilst these children are categorised as life-limiting they have distinct needs and trajectories.
9. Price et al. (2012)	To examine the experiences of	Qualitative.	25 parents (16 mothers and 9	In-depth interviews.	<ul style="list-style-type: none"> <li>• Although parents' accounts</li> </ul>	The findings revealed

Ireland	bereaved parents concerning the care provided to children who died from cancer compared to those who died from a non-malignant condition.		fathers) talked about the life and death of 16 children. 10 of the children had non-malignant life-limiting conditions while 6 had cancer.		<p>displayed commonalities, key differences were discernible.</p> <ul style="list-style-type: none"> <li>• Typically, parents of children with cancer considered that care at the end of life as well-resourced and responsive to their and their child's needs.</li> <li>• In contrast, parents of children with non-malignant conditions reported under-resourced and inadequately responsive services.</li> <li>• Although both groups of parents called extensively on military metaphors such as 'battle', 'fight' and 'struggle' the focus of their respective energies was different.</li> <li>• In the one case the adversary was disease and illness; in the other it was the service providers and service provision.</li> </ul>	significant issues regarding service provision for children with non-malignant life-limiting conditions.
10. Redmond and Richardson (2003) Ireland	Explore mothers' views of the usefulness of the financial, practical and emotional supports being	Largely qualitative.	17 mothers of children aged 4 years and under with severe/profound intellectual disability,	Semi-structured interviews. To allow for further perspectives in the research, certain	<ul style="list-style-type: none"> <li>• The study reveals these mothers to be engaged in stressful but skilled care of their children with a clear wish to continue</li> </ul>	The nature of the disabilities suffered by these young children was diverse, ranging from significant birth trauma

	offered to them and their suggestions for service improvements.		some with attendant complex medical, life-limiting conditions.	demographic features of the group of mothers were identified. The General Health Questionnaire (GHQ) (12) was also administered to mothers at the time of the interview.	<p>caring for their child in the family home.</p> <ul style="list-style-type: none"> <li>Mothers frequently referred to the process of gaining useful information on services as 'haphazard' and most of the services offered to them as uncoordinated, unreliable and difficult to access.</li> <li>The study reveals that many of these children's needs are not being adequately met by either the intellectual disability services or the acute medical services, and some families are forced to privately finance services such as physiotherapy and speech therapy.</li> </ul>	through serious metabolic disorders to complex congenital abnormalities. The severity of the disabilities and the medical complications experienced by many of these children adversely affected their long-term survival.
11. Smith, Graham and Herbert (2017) Australia	1. Clarify the respite needs of parents caring for children with life-limiting conditions by first identifying the respite, both formal and informal, that these families access and then identifying preferences for future respite.	Descriptive cohort survey.	34 families cared for by the Paediatric Palliative Care Service in Queensland were approached to participate in a survey. 20 surveys were returned.	20 question survey.	<ul style="list-style-type: none"> <li>3 of the families (15%) reported receiving no respite in the previous 12 months.</li> <li>Families who received respite received a combination of formal respite (a structured care provider) and informal respite (family or friends).</li> </ul>	The study was confined to one specific region.

	2. To understand barriers that families have to accessing the respite they need in caring for their child.				<ul style="list-style-type: none"> <li>• 10 (50%) reported that they would want the time of respite changed.</li> <li>• Barriers to receiving adequate respite included complexity of care of the child, financial barriers and lack of a respite provider.</li> </ul>	
12. Swallow, Forrester and Macfadyen (2012) United Kingdom	Explore teenagers and parents views on a short-break service for children with life-limiting conditions.	Qualitative.	15 mothers and 5 fathers (representing 15 families) of the 16 using the short-break service at the time. 5 teenagers (aged 14-18 years) with sufficient (but limited) ability, to participate individually.	Five individual interviews and four focus groups with parents, and two focus groups with teenagers.	<ul style="list-style-type: none"> <li>• Three themes emerged: accessibility and communication; needs and boundaries; and shaping the service.</li> <li>• Teenagers enjoyed regular planned residential breaks, access to skilled staff and bespoke facilities to support their needs, opportunities to meet others with life-limiting conditions and fun time away from home, thereby giving parents peace of mind, a regular planned break from care-giving, opportunities to meet other parents and to spend exclusive time with their other children.</li> </ul>	Of the 16 children who utilised the short-break service 11 children were unable to participate because of profound communication difficulties, but parents' accounts acted as a proxy. On average, families booked short breaks 10 times a year.
13. Weaver et al. (2018)	How the pediatric home nursing	Quantitative	A total of 38 home health nursing	A survey on their perceived	<ul style="list-style-type: none"> <li>• The gap in the average number of nursing hours</li> </ul>	The response rate was 63% (38/60).

<p>United States</p>	<p>shortage translates into a lived experience for families with children with complex medical conditions receiving palliative care.</p>		<p>surveys were completed by families receiving pediatric palliative care consultation services at a freestanding children's hospital in the Midwest.</p>	<p>home health nursing needs and their experiences accessing home health nursing. The survey was offered in English or Spanish.</p>	<p>allotted versus received was 40 hours per week per family, primarily during evening hours.</p> <ul style="list-style-type: none"> <li>• Parents missed an average of 23 hours of employment per week to provide hands-on nursing care at home, ranking stress regarding personal employment due to nursing shortage at 6.2/10.</li> <li>• Families invested an average of 10 hours per month searching for additional nursing coverage and often resorted to utilizing more than 6 different home nurse coverage personnel per month.</li> <li>• Families reported multiple delays to hospital discharges (mean, 15 days per delay) due to inability to find home nursing coverage.</li> <li>• Respiratory technology and lack of Medicaid coverage (<math>P &lt; .02</math>) correlated with the gap in home nursing access.</li> </ul>	<p>Limitations to this study include small sample size and one-site location.</p>
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Author, Year and Country	Aim/objective	Study design	Sample	Data collection techniques	Key findings	Comments
1. Allen (2014) United States	Describe possible factors that affect parental decision-making for medically complex children.	Integrated literature review.	PubMed, Cumulative Index of Nursing and Allied Health Literature (CINAHL), and PsycINFO were searched using the combined key terms 'parents and decision-making' to obtain English language publications from 2000 to June 2013.	The search strategy generated 336 articles relevant based on their titles with 305 articles eliminated after review of abstract.	<ul style="list-style-type: none"> <li>The strengths of the empirical research reviewed are that decisions about initiating life support and withdrawing life support have received significant attention.</li> <li>Researchers have explored how many different factors impact decision-making and have used multiple different research designs and data collection methods to explore the decision-making process.</li> <li>These initial studies lay the foundation for future research and have provided insight into parental decision-making during times of crisis.</li> </ul>	The critical decisions included continuation or termination of a high-risk pregnancy, initiation of life-sustaining treatments such as resuscitation, complex cardiothoracic surgery, use of experimental treatments, end-of-life care, and limitation or withdrawal of support.
2. Beecham et al. (2017) United Kingdom	Investigate how parents of children with life-limiting conditions approach and experience advanced care planning.	Qualitative.	Parents of 18 children; 9 children who were currently receiving palliative care services, and 9 children who had received palliative	Open-ended, semi-structured interviews. In 6 cases, parents completed one interview, in 11 cases parents completed two interviews and in 1 case parents	<ul style="list-style-type: none"> <li>Parents reported having discussions and making decisions about the place of care, place of death and the limitation of treatment.</li> <li>Most decisions were made relatively late in</li> </ul>	All parents were offered a follow-up interview. The majority of the children had non-malignant life-limiting conditions.

			care and died.	completed three interviews at the family's request.	<p>the illness and by parents who wished to keep their options open.</p> <ul style="list-style-type: none"> <li>Parents reported different levels of involvement in a range of decisions; many wished to be involved in decision making but did not always feel able to do so.</li> </ul>	
3. Caeymaex et al. (2013) France	Investigate whether parental perception of the type of involvement in the end-of life decision-making process for their child in the NICU is related to their long-term grief outcome.	A retrospective study with mixed methods.	78 parents of 53 children who died in NICU.	Face to face interviews. Grief reactions were assessed with the Texas Revised Inventory of Grief.	<ul style="list-style-type: none"> <li>In decisions to forego life-sustaining treatment in the NICU, the perception of a shared decision is associated in the long term with lower grief scores than perceptions of the other types of decision-making.</li> </ul>	The children died in in one of four NICUs in different areas in France.
4. Carroll et al. (2012) United States	Explore influences on decision making identified by parents of children receiving pediatric palliative care.	Part of a mixed method study.	73 parents of 50 pediatric patients who were referred to the hospital's pediatric palliative care service.	Semi-structured interviews.	<ul style="list-style-type: none"> <li>Four dominant interrelated themes permeated parents' discussions about the decisions they were making for their children and the process of decision making.</li> <li>First, Orientation and Direction connotes the parents' effort to establish and clarify the broad context of decision making.</li> </ul>	The authors concluded that parents report grappling with several influences upon their decision-making processes that extend well beyond the standard discussions of medical information exchanges and the evaluation of risks and benefits.

					<ul style="list-style-type: none"> <li>• Second, Defining What Is Good for the Child conveys how the parents posed questions and pondered what decisions would be in the child's best interests.</li> <li>• Third, the entwined theme of Relationships, Communication, and Support reflects how parents reported the social and interactive nature of decision making.</li> <li>• Fourth, the theme of Feelings and Personal Accountability focuses inward as parents report efforts to deal with their emotional responses.</li> </ul>	
5. Dellon et al. (2010) United States	Describe caregivers' perspectives on decision making for the use of intensive treatments for patients with advanced cystic fibrosis (CF) lung disease.	Qualitative.	36 caregivers of 36 patients who died of CF.	Semi-structured interviews.	<ul style="list-style-type: none"> <li>• Twenty (56%) patients received intensive treatments during the last week of life.</li> <li>• Twenty-two (61%) caregivers reported ever having discussed intensive treatment preferences with a physician, and 17 (77%) of these discussions were initiated during an acute illness.</li> </ul>	The majority of the patients who died from CF were adults, however 12 of the patients were under eighteen.

					<ul style="list-style-type: none"> <li>• Only 14 (39%) of all patients participated.</li> <li>• Caregivers expressed less certainty about consistency of treatments with patient preferences when patients did not participate.</li> <li>• Twenty-nine (81%) caregivers endorsed first discussing treatment preferences during a period of medical stability.</li> </ul>	
6. Eden and Callister (2010) United States	Evaluate parental involvement in end-of-life care and decision making for their infant in the newborn intensive care unit.	Integrated literature review.	An electronic search was undertaken to identify studies that included end-of-life care published between 2000 and 2008 in the following databases: CINAHL, Alt Health Watch, Applied Science & Technology Abstracts, Health Source - Consumer Edition and Academic Edition, MEDLINE, PsycARTICLE S, PsycCRITIQU	A total of 10 studies were included.	<ul style="list-style-type: none"> <li>• Findings revealed that establishing good relationships and clear communication between health-care providers and parents builds trust and eases stress placed on parents making decisions about the care of their infant.</li> <li>• Palliative care programs provide support for parents and facilitate their decision making.</li> <li>• Parents can be educated about how to communicate with health-care providers.</li> <li>• Educating nurses on how to provide end-of-life care may also</li> </ul>	The authors concluded that as advances in technology preserve the lives of very low birth weight and critically ill newborn infants in the NICU, ethical dilemmas regarding treatment become more complex. Parents need to be closely involved in the care and decision-making process associated with their infant.

			ES, PsycINFO, and The Cochrane Library.		help improve support for parents during this difficult time.	
7. Geurtzen et al. (2017) Netherlands	Evaluate Decisional Conflict and Regret among parents regarding the decision on initiating comfort or active care in extreme prematurity and to relate these to decision-making characteristics.	Nationwide, multicentre, cross-sectional study.	61 parents of infants born at 24-27 weeks gestational age.	Online survey. The survey contained a Decisional Conflict and Decision Regret Scale.	<ul style="list-style-type: none"> <li>The median Decisional Conflict score was 28.</li> <li>From the subscores within Decisional Conflict, 'values clarity' revealed the highest median score of 42- revealing that parents felt unclear about personal values for benefits and risks of the decision on either comfort care or active care.</li> <li>The median Decision Regret score was 0.</li> <li>Regret scores were influenced by the actual decision made and by the outcome.</li> <li>Decision Regret was lower in the active care group and the survivor group.</li> </ul>	This study was included as the infants were 24-27 weeks gestational age. Most doctors define the age of viability as being about 24 weeks of gestation.
8. Knapp et al. (2014) United States	Investigate how decisional conflict varies among racial and ethnic subgroups.	Quantitative.	266 parents whose children have a life-threatening illness.	Telephone surveys were conducted in English and in Spanish. To measure decisional conflict and its components the Decisional Conflict Scale was used.	<ul style="list-style-type: none"> <li>Bivariate results suggest that minority parents report less Effective Decision Making (<math>p &lt; 0.05</math>) and report less Support in Decision Making (<math>p &lt; 0.05</math>) compared to</li> </ul>	Gender of child, age of child, and parents rating of child's health (ranging from 'excellent' to 'poor') are included in the analysis. Additional factors, including

				<p>The impact a child's illness has on the family was measured using the Impact of Family Scale.</p>	<p>white, non-Hispanic parents.</p> <ul style="list-style-type: none"> <li>• For the subgroup analysis, those who identify as Mexican American and Central/South American report having greater Uncertainty in Choosing Options (<math>p &lt; 0.05</math>) and less Values Clarity (<math>p &lt; 0.05</math>) as compared to Puerto Rican or Cuban Americans.</li> <li>• Results from the multivariate analyses suggest that those whose primary language is not English are associated with greater Uncertainty in Choosing Options (<math>p &lt; 0.05</math>).</li> <li>• Values Clarity was lower for children who were diagnosed with their life-threatening condition at birth (<math>p &lt; 0.05</math>) as compared to children diagnosed at a later time.</li> </ul>	<p>primary language in the home, parent/caregiver's relationship to the child (parent or nonparent), marital status of parent/caregiver, household type, educational level of parent/caregiver, and age of the parent/caregiver were also used in the analyses.</p>
<p>9. Ling et al. (2015) Ireland</p>	<p>Examine the views and experiences of parents of children with life-limiting conditions on out-of-home respite care and to present a</p>	<p>Multiple, longitudinal, qualitative case study design.</p>	<p>Parents (9 mothers and 3 fathers) of children with life-limiting conditions.</p>	<p>Multiple, in-depth interviews. A total of 19 in-depth interviews were conducted.</p>	<ul style="list-style-type: none"> <li>• Parents reported vastly different needs and experiences of respite.</li> <li>• Cross-case comparison showed that for all parents utilising respite</li> </ul>	<p>The title of this study refers to decision making; however the study also dealt with issues surrounding access to respite care.</p>

	proposed new model of care based on the findings.				<p>care, regardless of their child's age and condition, home was the location of choice.</p> <ul style="list-style-type: none"> <li>• Many interlinking factors influenced their decisions on utilisation of out-of-home respite care including: past experience of in-patient care, and trust and confidence in care providers.</li> <li>• Issues were raised regarding the impact of care provision in the home on family life, siblings and the concept of home.</li> </ul>	Pertinent matters regarding decision-making on utilisation of out-of-home respite care were explored in this study. These were highlighted in the study findings. This study was also valuable in examining parents' experiences.
10 Michelson et al. (2009) United States	Explore factors described by parents in the pediatric intensive care unit (PICU) as important/influential if they were to consider withdrawing life-sustaining therapies.	Qualitative.	70 parents whose child was admitted to the PICU for more than 24 hours to up to 1 week.	Semi-structured one-to-one interviews.	<ul style="list-style-type: none"> <li>• Parents describe a broad range of views regarding possible consideration of withdrawing life-sustaining therapies for their children and what factors might influence such a decision.</li> </ul>	The authors acknowledge the main study limitation as asking parents hypothetical questions. Parents' reactions when confronting imminent end-of-life decisions could differ.
11. Nicholson (2012) United Kingdom	To investigate parents and young people's experiences of making decisions about technological support, focusing on artificial nutrition and assisted ventilation.	Qualitative.	19 families of life-limited children and young people (25 parents and young people) who had considered the use of technological support was	In-depth interviews.	<ul style="list-style-type: none"> <li>• Parents and young people wish to make the right choices, and consider a range of factors and information in an effort to become informed, even when there is no perceived choice in decisions they make.</li> </ul>	This study was part of a thesis. The study site was confined to one children's hospice. As a result, it is possible that the study has not been able to capture the full range and diversity of

			drawn from a children's hospice.		<ul style="list-style-type: none"> <li>Quality of life is identified as the key decision factor, which as a fluid and evolving concept underpins the process of assessing a child's needs and considering the potential impact of a proposed intervention.</li> </ul>	views on the decision process around the use of technological support.
12. Popejoy (2015) United Kingdom	To understand the 'lived experience' of parents throughout the process of making and revising end of life care decisions for their child.	Qualitative.	3 mothers who had been bereaved as a result of their child's life-limiting illness.	One-to-one semi-structured interviews.	<ul style="list-style-type: none"> <li>Two overarching themes were identified: making decisions and revising and implementing end of life care plans.</li> <li>Sub-themes included: who should be the decision maker; when discussions should be initiated; the values underpinning the plans; revisiting the plans; and barriers and facilitators to their implementation.</li> <li>Parents understand the importance of planning for the end of their child's life but find the process difficult.</li> <li>They also find it a challenge to verbalise their decisions at the end of their child's life and</li> </ul>	The number of mothers who participated was very small. All three children had Category 3: progressive conditions without curative treatment options.

					value having the decision partly taken away from them.	
13. Rapoport et al. (2013) Canada	Explore parental perceptions of forgoing artificial nutrition and hydration during end-of-life care.	Qualitative.	Parents whose children died after a decision to forgo artificial nutrition and hydration during end-of-life. A total of 11 parents from 6 families (5 fathers, 6 mothers) participated.	In-depth interviews.	<ul style="list-style-type: none"> <li>All parents were satisfied with their decision to forgo artificial nutrition and hydration and believed that their child's death was generally peaceful and comfortable.</li> <li>The child's perceived poor quality of life was central to the decision to forgo artificial nutrition and hydration, with feeding intolerance often contributing to this perception.</li> <li>Despite overall satisfaction, all parents had doubts and questions about the decision and benefitted from ongoing assurances from the clinical team.</li> </ul>	The authors concluded that forgoing artificial nutrition and hydration in children at end-of-life is an acceptable form of palliation for some parents and may contribute to a death that is perceived to be peaceful and comfortable. In situations which forgoing artificial nutrition may be a reasonable possibility, physicians should be prepared to introduce the option.
14. Tamburro et al. (2011) United States	Describe goals of care for children with complex, life-limiting conditions and to assess the variables that may influence these goals.	Quantitative.	50 children (ages ranged from newborn up to 19 years with a median age of 4.6 years) and their families. 37 children had significant cognitive delay/impairment.	Goals of care were elicited from the parents and children with complex life-limiting conditions during initial palliative care consultation. These were categorized into one of four quality-of-	<ul style="list-style-type: none"> <li>One hundred and forty goals of care were obtained from 50 children/parents.</li> <li>49 children/parents identified at least one goal pertaining to physical health and independence.</li> <li>This was significantly more</li> </ul>	The authors concluded that children with life-limiting conditions and their families commonly verbalize goals related to health maintenance and independence. Anticipating this may foster

			Neuromuscular disorders accounted for more than half of the diagnoses.	life domains: 1) physical health and independence, 2) psychological and spiritual, 3) social, and 4) environment.	<p>than any other category (<math>p &lt; 0.0001</math>).</p> <ul style="list-style-type: none"> <li>33 of the 50 children/parents opted for full medical support at the time of initial consult (time from diagnosis until palliative care consult ranged widely from less than a month to 19 years with a median of 21.1 months).</li> </ul>	communication and improve patient care. This is also very significant regarding decision-making.
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Author, Year and Country	Aim/objective	Study design	Sample	Data collection techniques	Key findings	Comments
1. Baverstock and Finlay (2008) United Kingdom	To describe how paediatric consultants' report dealing with child and neonatal deaths as part of their daily work.	Mixed method.	61 experienced consultants (40 male and 21 female).	Pre-piloted self-administered questionnaire	<ul style="list-style-type: none"> <li>• Most consultants had experienced a variety of opinions when talking to parents about when to withdraw or withhold life sustaining treatment.</li> <li>• Uncertainty (39%) and disagreement (44%) made decision making difficult.</li> <li>• Discussion with colleagues (41%), 'honesty and time with parents' (28%) and planning or 'stage managing' talking with parents (38%) were well used strategies to deal with this.</li> <li>• New consultants find responsibility and decision making the biggest change from being a registrar.</li> <li>• Many consultants felt it important to have their own support network.</li> </ul>	The findings also reveal pertinent information regarding the complexity of decision-making. Included consultants working in a variety of specialties. It was difficult to interpret the percentage of consultants involved who cared for children with non-malignant life-limiting conditions. Confined to one specialty.
2. Beaune et al. (2014) Canada	Examine the content validity and utility for health care providers of a poverty screening tool and companion resource guide	Mixed method.	8 health care providers (n=8) including 3 physicians, 4 nurses, and 1 social work intern.	The quantitative component utilized a content validity measure to address the validity, relevance, and feasibility of the	<ul style="list-style-type: none"> <li>• The study found high relevance and validity of the tool.</li> <li>• Significant barriers to implementing the screening tool in clinical practice</li> </ul>	There are several limitations to this study. For example, the participants may not be representative of all pediatric

	for the pediatric palliative care population.			Child Poverty Tool and resource guide. The qualitative component of the study consisted of a focus group with the same health care providers.	were described by participants, including: concerns regarding time required, roles and responsibilities, and discomfort in asking about income.	palliative care professionals.
3. Bergstraesser et al. (2013) Switzerland	Describe the perceptions and needs of pediatric health care professionals taking care of children with palliative care needs and to develop a concept for the first Center of Competence for Pediatric Palliative Care.	Qualitative.	76 healthcare professionals , including physicians, nurses, psychologists and social workers.	Semi-structured interviews.	<ul style="list-style-type: none"> <li>Healthcare professionals expressed openness to pediatric palliative care and reported distinctive needs for support in the care of these patients.</li> <li>The main tasks of specialized pediatric palliative care teams in Switzerland would encompass the coaching of attending teams, coordination of care, symptom control, and direct support of affected families during and beyond the illness of their child.</li> </ul>	This study indicates the need for specialized pediatric palliative care in Switzerland. Specialized pediatric palliative care teams could have a significant impact on the care of children and families with pediatric palliative care needs. Whether hospices are an option in Switzerland remains unanswered; however, a place to meet other families with similar destinies was emphasized.
4. Clarke and Quin (2007) Ireland	Explore professional carers' experiences of providing a pediatric palliative care service in Ireland.	Qualitative.	15 participants. The participants included doctors, nurses, physiotherap	Focus group interviews.	<ul style="list-style-type: none"> <li>Three theoretical themes included clarity of definition and complexity of engagement, seeking to deliver a palliative care service, and the emotional cost of</li> </ul>	This qualitative study was part of a national pediatric palliative care needs analysis.

			ists, occupational therapists and social workers.		<p>providing palliative care.</p> <ul style="list-style-type: none"> <li>• Further analysis of these themes points to a work-life experience of skilled and emotional engagement with children, and their parents, in complex processes of caregiving and decision making.</li> <li>• Pediatric palliative care occurs in an environment where parents shoulder a large burden of the care and professionals find themselves working in under resourced services.</li> </ul>	Professionals were involved in care of children with a range of life-limiting conditions. Researcher reflexivity and context not addressed.
5. Dryden-Palmer et al. (2018) Canada	Describe the consequences of workplace stressors on healthcare clinicians in PICU, and strategies for personal well-being, and professional effectiveness in providing high-quality end-of-life care.	Literature review, clinical experience, and expert opinion.	A sampling of foundational and current evidence was accessed.	Narrative review and experiential reflection.	<ul style="list-style-type: none"> <li>• The well-being of clinicians influences the quality and effectiveness of patient care, team functioning, and the retention of skilled individuals in the PICU workforce.</li> <li>• End-of-life care, including decision making, can be complicated.</li> <li>• Both are major stressors for PICU staff that can lead to adverse personal and professional consequences.</li> <li>• Overresponsiveness to routine stressors may be seen in those with moral distress.</li> <li>• Underresponsiveness may be seen in those with compassion fatigue or burnout.</li> </ul>	The authors concluded that a creative, comprehensive, and sustained approach to the stressors inherent in the PICU can help assure that individuals and teams are at their best to provide effective, end-of-life care for children and their families.

6. Forster and Haiz (2015) Australia	To explore health professionals' perceptions of bereavement support surrounding the loss of a child.	Qualitative.	10 health professionals including doctors, nurses and social workers who were directly involved in the care of the dying child and family in 7 cases of paediatric death in medical, oncology and paediatric intensive care at the tertiary paediatric hospital.	Semi-structured interviews.	<ul style="list-style-type: none"> <li>• For health professionals, constructions around coping emerged as peer support, personal coping strategies, family support, physical impact of support and spiritual beliefs.</li> <li>• Analysis of the narratives also revealed health professionals' perceptions of their support provision.</li> </ul>	The diagnoses were not revealed. It was unclear as to how many (if any) of the children had non-malignant life-limiting conditions.
7. Lee and Dupree (2008) United States	Describe the experiences of pediatric intensive care unit (PICU) staff caring for a child who dies, and to determine whether responses included unprompted indications of moral distress as described in the literature.	Qualitative descriptive.	29 staff members regarding 8 patient deaths.	Semi-structured interviews.	<ul style="list-style-type: none"> <li>• The major themes of staff members experiences were (1) importance of communication, (2) accommodating the wishes of others despite personal preferences, (3) ambiguity about the use of technology, (4) sadness, and (5) emotional support.</li> <li>• Descriptions of moral distress were seen infrequently.</li> <li>• Many welcomed the sadness they experienced as a sign of their humanity and</li> </ul>	This study was conducted in a large PICU. Participants included nurses, physicians, and psychosocial support personnel.

					emotional availability, but did not feel adequately supported in dealing with their grief.	
8. Llewelyn et al. (2015) United Kingdom	Understand how healthcare professionals conceptualise spirituality among seriously ill children and young people.	Report of a workshop.	Healthcare professionals in the community caring for children with life-limiting conditions.	Presentations and small group discussions.	<ul style="list-style-type: none"> <li>Healthcare professionals conceptualised spirituality as highly individualised searches for meaning, hope and connectedness to self, others and the world.</li> <li>They saw spirituality within a development context.</li> <li>Healthcare professionals described spiritual concerns that were tied to their own conceptualisations of spirituality, centring on ideas of loss, including loss of hope or meaning.</li> <li>Healthcare professionals approached spiritual concerns of children/young people by 'being there' and supporting spiritual enquiry.</li> <li>Challenges to their work included managing hopes of children/young people and families in the</li> </ul>	The workshop took place in a large paediatric palliative care network.

					<p>face of poor prognosis, discussions about miracles and issues with their own faith.</p> <ul style="list-style-type: none"> <li>• Spiritual care was seen as different to other areas of care which healthcare professionals felt had a greater prescription in delivery.</li> </ul>	
9. McCloskey and Taggart (2010) Ireland	Explore the experiences of stress in nurses providing children's palliative care in one region of the United Kingdom.	Qualitative.	Children's hospice nurses, community children's nurses and children's nurse specialists based in the regional children's hospital. In total 18 nurses participated.	Focus groups using semi-structured interviews.	<ul style="list-style-type: none"> <li>• Four core themes emerged: work demands; relationships, maintaining control, and support and roles.</li> <li>• Of particular note were stressors associated with the sub-themes of relationships, emotional demands and ethical conflicts.</li> </ul>	The nurses were recruited through a purposeful sampling approach from the total population of all children's palliative care nurses working within three settings across Northern Ireland (n=75). The overall response rate to the invitation to participate in the study was 56% but scheduling difficulties due to sick leave, work commitments and annual leave reduced the final response to 24%.
10. McConnell and Porter (2017)	Explore the experiences of care team staff	Qualitative.	15 care team staff from a children's	One-to-one interviews and focus groups.	<ul style="list-style-type: none"> <li>• The hospice setting provides a model of</li> </ul>	Twelve individual interviews and

Ireland	who provide end-of-life care within a children's hospice.		hospice offering palliative and specialist care to life-limited children and young people.		excellence in supporting staff and mitigating challenging aspects of their role, which includes peer/organisational support, and regular ongoing training in key aspects of children's palliative care.	one focus group with three participants were conducted, involving eight staff nurses, two healthcare assistants, one senior healthcare assistant, and one care team manager. Researcher reflexivity not addressed.
11. McConnell, Scott and Porter (2016) United Kingdom	Explore the experiences of healthcare professionals who provide end-of-life care to children in order to inform the development of interventions to support them.	A mixed-method review.	Searches included CINAHL, MEDLINE, Web of Science, EMBASE, PsycINFO and the Cochrane Library in June 2015, with no date restrictions. Additional literature was uncovered from searching reference lists of relevant studies, along with contacting experts in the field of paediatric palliative care.	A total of 30 studies were included. This included 16 qualitative, 6 quantitative and 8 mixed-method studies.	<ul style="list-style-type: none"> <li>Key themes identified rewards and challenges of providing end-of-life care to children, the impact on staff's personal and professional lives, coping strategies and key approaches to help support staff in their role.</li> </ul>	This was a systematic mixed studies review. Study selection, appraisal and data extraction were conducted by two independent researchers. Integrative thematic analysis was used to synthesise the data.

12. Pontin and Lewis (2008) United Kingdom	Explore how community children's nurses deliver services to children with life-limiting, life-threatening and chronic conditions.	Qualitative-action research study.	The co-inquirers were a children's nurse lecturer, a senior community children's nurse, and 16 co-inquiring community children's nurses who provide services to children living with life-limiting, life-threatening, and chronic conditions.	In-depth interviews.	<ul style="list-style-type: none"> <li>• The study generated a number of insights about the sorts of phenomena that contribute to community children's nurses perceptions of workload.</li> <li>• Themes included strategy, being proactive, purposeful visit, and knowing families.</li> </ul>	Confined to one professional group. A number of insights about the sorts of phenomena that contribute to community children's nurses perceptions of workload were generated.
13. Quinn and Bailey (2011) Ireland	Explore the experiences of Irish palliative care clinical nurse specialists caring for children and families in the community.	Qualitative.	7 community palliative care clinical nurse specialists from across one health region in the Republic of Ireland.	Focus group.	<ul style="list-style-type: none"> <li>• Four key themes emerged: gaining access to the child and family, role complexities, pressures of caring, and support strategies.</li> <li>• Provision of community children's palliative care by the clinical nurse specialist is complex.</li> <li>• The participants demonstrated their commitment to consult, coordinate, negotiate, and ultimately deliver the care required by children and families, but</li> </ul>	The number of clinical nurse specialists that participated was small. Issues of access were also raised. Again the study was limited to one region.

					against a background of issues relating to accessing the patient and family, the complexity of the role, and the pressure that such work incurs.	
14. Reid (2013) Scotland	Elicit the views of children's nurses with regard to the personal, contextual and interprofessional challenges faced when delivering palliative and end of life care to children and young people in the community.	Qualitative.	7 nurses (4 children's community nurses and 3 children's respite nurses) who provide palliative care to one or more child or young person in the home.	Semi-structured interviews.	<p>Four themes emerged:</p> <ul style="list-style-type: none"> <li>• Service delivery: one of the main challenges identified by all participants, especially when there was rapid turnover of staff, was initiating and sustaining plans of care across a range of contexts.</li> <li>• Nurse-family relationships: maintaining professional boundaries when trying to balance familiarity and emotional involvement with a level of detachment was challenging.</li> <li>• Nurses' grief: although participants appeared to be reconciled to child death, all still described strong feelings of fearful anticipation, emotional wrestling and cumulative effects.</li> <li>• Funeral rites and bereavement</li> </ul>	Limited to one professional group. Pertinent information regarding nurses' experiences. Issues of access were likewise revealed in the study findings. Nurses experienced considerable internal and external pressures. Some are inevitable but others, such as organisation of care provision to families and nurses' personal coping, could be improved by adequately resourced workforces, integrated service structures and guidance on reflective practice.

					support: the funeral appeared to offer a setting where most nurses could legitimately demonstrate personal grief.	Researcher reflexivity not addressed.
15. Taylor and Aldridge (2017) United Kingdom	Explore the rewards and challenges of working in a children's hospice with an aim to identify staff support and development needs.	Qualitative exploratory design.	34 care team members including medical, nursing and allied health professionals participated in an interview. 17 care team members took part in focus groups. 3 care team members took part in both the interview and focus group.	Face-to-face interviews and focus groups. Focus groups comprised of approximately six care team members.	<ul style="list-style-type: none"> <li>• Participants described the work as emotionally intensive and multi-faceted.</li> <li>• 'Getting it right' was identified as a strong motivator and reward, but also a potential stressor.</li> <li>• Other factors were identified as both a reward and stressor, including team functioning, the allocation of work, meeting parent expectations, and the environment.</li> <li>• Participants identified training needs to help them feel more confident and competent.</li> <li>• They expressed concerns about work-related stress.</li> <li>• Informal support and group clinical reflection were identified as primary resources. However, opportunities for this were limited.</li> </ul>	As well as varying in occupational background and professional training the participants had worked at the hospice for different durations, ranging from 10 months to more than 20 years.

Author, Year and Country	Aim/objective	Study design	Sample	Data collection techniques	Key findings	Comments
1. Delany et al. (2017) Australia	Ascertain clinicians' views on the acceptability and usefulness of a handbook and web-based resource (Caring Decisions) that was designed as an aid for parents facing end-of-life decisions for their child.	Qualitative.	18 healthcare professionals who were closely involved in clinical decision-making and/or provision of care to parents of children who face life-limiting illnesses.	One-to-one semi-structured interviews.	<ul style="list-style-type: none"> <li>Data analysis confirmed the acceptability and usefulness of the resource.</li> </ul> <p>Two major themes were revealed:</p> <ul style="list-style-type: none"> <li>Family empowerment, with sub-themes <i>Giving words and clarity</i>, <i>Conversation starter</i>, <i>'I'm not alone in this'</i>, and <i>A resource to take away</i>, highlighted how the resource filled a gap by supporting and enabling families in a multitude of ways;</li> <li>Not just for families, with sub-themes <i>A guide for staff</i>, <i>When to give the resource?</i>, <i>How to give the resource</i> and <i>Who should give the resource?</i>, explored the significant finding that participants viewed the resource as a valuable tool for themselves, but its presence also brought into relief potential gaps in communication</li> </ul>	The findings from this study also provide valuable insight regarding decision-making.

					processes around end-of-life care.	
2. Heath et al. (2012) United Kingdom	Explore the views of stakeholders on providing paediatric 'care closer to home', in community-based outpatient clinics delivered by consultants.	Qualitative.	37 stakeholders including healthcare professionals, managers, commissioners and executive team members.	Semi-structured interviews.	<ul style="list-style-type: none"> <li>Participants acknowledged that outreach clinics would involve a change in traditional ways of working and that the physical setting of the clinic would influence aspects of professional practice.</li> <li>Different models of 'care closer to home' were discussed, as were alternatives for improving access to specialist care.</li> <li>Participants supported 'care closer to home' as a good principle for paediatric outpatient services; however the challenges of setting up and maintaining community clinics meant they questioned how far it could be achieved in practice.</li> </ul>	Although not specifically related to paediatric palliative care this population is included. The authors concluded that the place of service delivery is both an issue of physical location and professional identity. Policy initiatives which ignore assumptions about place, power and identity are likely to meet with limited success.
3. Hoell et al. (2017) Germany	First-to determine the number of children in pediatric palliative care without a clear diagnosis—second—to determine the clinical	Single-centre retrospective cohort study.	27 children without a clear diagnosis or brain injured at the start of pediatric palliative home care.	Patient data were routinely entered into a web interface database by members of the pediatric palliative care team and from there extracted and	<ul style="list-style-type: none"> <li>Children without a clear diagnosis are relatively common in pediatric palliative care and have—like all other patients—the right to receive optimized and symptom-</li> </ul>	The findings from this study reveal important matters concerning access to services and decision-making.

	characteristics and symptoms, supportive needs, and the process of decision-making in those, and—third—to compare these data with the data of children with similar signs and symptoms but a confirmed diagnosis.			further processed. Information included demographic data, number of home visits, symptoms, medications, reasons for referral to the pediatric palliative care team, and information on advance care decision-making.	<p>adapted palliative care.</p> <ul style="list-style-type: none"> <li>• Parents are less likely to choose treatment limitation for children who lack a definitive diagnosis.</li> </ul>	
4. Hoell et al. (2017) Germany	Develop an empirically based conceptual model of the needs of children with life-limiting conditions.	Concept mapping methodology	50 healthcare professionals with expertise in pediatric palliative and hospice care.	Brainstorming on children's needs. Sorting and rating the needs statements.	<ul style="list-style-type: none"> <li>• The seven-cluster concept map appeared best in terms of both interpretability and parsimony. This model includes the following clusters of needs: 1) pain, 2) decision-making, 3) medical system access and quality, 4) dignity and respect, 5) family-oriented care, 6) spirituality, and 7) psychosocial issues.</li> </ul>	The findings reveal pertinent issues regarding children's needs and service provision. The importance of decision making was also highlighted.
5. Price, Jordan and Prior (2013) Ireland	To investigate health and social care professionals' perspectives on developing services for children with life limiting conditions at the end-of-life stage using	Qualitative.	35 health and social care professionals (the study participants included doctors, nurses, healthcare assistants	Focus groups.	<p>Six issues were identified across professional groupings as particularly challenging:</p> <ul style="list-style-type: none"> <li>• truth telling</li> <li>• symptom management</li> <li>• communication with, and relationships between families and professionals</li> </ul>	Differences in the issues identified as problematic and priorities for service development emerged between focus groups. The study took place in one

	<p>issues identified by bereaved parents as priorities.</p>		<p>and play specialists).</p>		<ul style="list-style-type: none"> <li>• emotional impact</li> <li>• the withdrawal of feeding or treatment and</li> <li>• sibling support</li> </ul> <p>Collective findings demonstrated resonance between priorities identified by parents from an earlier study (Price et al., 2011; Price et al., 2012). Conversely, late referral to hospice care and lack of services in the community dominated accounts of parents whose children had non-malignant conditions, but were not prioritised by professionals.</p>	<p>region in the UK.</p>
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Author, Year and Country	Aim/objective	Study design	Sample	Data collection techniques	Key findings	Comments
1. Cardona (2017) United States	Describe how a pediatric psychologist, who is integrated into an ethics committee, can promote joint decision-making, as well as serve in an important consultative role to medical teams to facilitate collaborative and compassionate care.	Case illustration.	A child diagnosed with a life-limiting condition at 5 months.	Case illustration.	<ul style="list-style-type: none"> <li>• The case provides an illustration of the many ethical challenges inherent in caring for profoundly medically ill children with life-limiting conditions.</li> <li>• Interdisciplinary care that is informed by ethical principles related to best interests of the patient, while keeping in mind the important psychological meaning that medically fragile children bring to their families, is critical to compassionate medical decision making.</li> <li>• Ethics consultations can assist teams in resolving disagreements regarding the goals of care, in addition to providing a model for open communication and collaborative problem solving.</li> </ul>	This case illustration demonstrates the complexity of decision-making for both parents/families and healthcare professionals.

2. De Vos et al. (2011) Netherlands	Investigate how Dutch pediatric specialists reach end-of-life decisions, how they involve parents, and how they address conflicts.	National cross-sectional survey.	136 pediatric specialists (including pediatric intensivists, oncologists, neurologists, neurosurgeons , and metabolic pediatricians) practicing in the 8 Dutch university hospitals.	Survey.	<ul style="list-style-type: none"> <li>• All responding physicians generally discuss an end-of-life decision with colleagues before discussing it with parents.</li> <li>• In half of the reported cases, respondents informed parents about the intended decision and asked their permission.</li> <li>• In one-quarter of the cases, respondents informed parents without asking for their permission.</li> <li>• In the remaining one-quarter of the cases, respondents advised parents and consequently allowed them to have the decisive voice.</li> <li>• The chosen approach is highly influenced by type of decision and type and duration of treatment.</li> <li>• Conflicts within medical teams arose as a result of uncertainties about prognosis and treatment options.</li> </ul>	Within Dutch pediatrics, end-of-life decisions are team decisions. Main conflict-solving strategies are taking more time and extending discussions.
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					<ul style="list-style-type: none"> <li>• Most conflicts arose because parents had a more positive view of the prognosis or had religious objections to treatment discontinuation.</li> <li>• All conflicts were eventually resolved by a combination of strategies.</li> </ul>	
3. Donnelly et al. (2005) United States	<p>1. Assess how directors of pediatric home ventilation programs facilitate shared decision-making with families facing decisions of whether to initiate or forgo long-term ventilation for their children with life-limiting conditions.</p> <p>2. Assess directors' perspectives on these families' decisional needs.</p>	Qualitative.	15 experienced physician directors.	Semi-structured interviews.	<ul style="list-style-type: none"> <li>• Physicians inform families of the potential benefits and burdens/risks of long-term ventilation for the child and of the option to forgo long-term ventilation.</li> <li>• They stress to families the physical, emotional, and social impact of caring for a child using long-term ventilation.</li> <li>• They recommend that decision-making around long-term ventilation should be interdisciplinary, initiated early, and not rushed.</li> <li>• They recommend that providers be transparent, candid, active</li> </ul>	Confined to one professional group.

					<p>listeners, and supportive.</p> <ul style="list-style-type: none"> <li>• They believe that the family's decision <b>should</b> be respected.</li> <li>• They vary in the extent to which they recommend an option to families.</li> <li>• They described barriers to decision-making that stem from families, providers, and other sources.</li> </ul>	
4. Edwards et al. (2012) United States	To discuss how best to conduct advanced care planning for children with life-limiting conditions on long-term assisted ventilation.	Retrospective study.	Retrospective analysis of end-of-life discussions regarding 47 deceased ventilator assisted children between 1987 and 2009.	Retrospective analysis of end-of-life discussions.	<ul style="list-style-type: none"> <li>• A total of 34 (72 percent) of 47 study patients were the subject of these discussions; many discussions occurred after acute deterioration.</li> <li>• They resulted in directives to forgo or limit interventions for 21 children (45 percent).</li> <li>• The authors surmise that many families were hesitant to discuss end-of-life issues during periods of relative stability.</li> </ul>	Included children with a wide range of life-limiting conditions including spinal muscle atrophy, neurodevelopmental delay and chromosomal abnormality.
5. Lotz et al. (2016) Germany	Investigate which factors paediatricians apply in	Qualitative.	17 experienced paediatricians from different	Focus groups.	<ul style="list-style-type: none"> <li>• The decisions about medical indication were based on</li> </ul>	Confined to one professional group.

	deciding about medical indication, and how they manage conflicts with parents.		specialties who were involved in the care of severely ill children/ adolescents (including children with life-limiting conditions).		<p>considerations relating to the individual patient, to the family, and to other patients.</p> <ul style="list-style-type: none"> <li>• Individual patient factors included clinical aspects and benefit–burden considerations.</li> <li>• Physicians’ individual views and feelings influenced their decision-making.</li> <li>• Different factors were applied or weighed differently in the two cases.</li> <li>• In case of conflict with parents, physicians preferred solutions aimed at establishing consensus.</li> </ul>	
6. Mitchell and Dale (2015) United Kingdom	Explore the experiences of senior medical and nursing staff regarding the challenges associated with Advanced Care Planning in relation to children and young people with life-limiting illnesses in the Paediatric Intensive Care Unit.	Qualitative.	8 Paediatric Intensive Care Unit consultants and 6 senior nurses participated.	Semi-structured interviews.	<p>Four main themes emerged:</p> <ul style="list-style-type: none"> <li>• recognition of an illness as 'life-limiting';</li> <li>• Advance Care Planning as a multi-disciplinary, structured process;</li> <li>• the value of Advance Care Planning and</li> <li>• adverse consequences of inadequate Advance Care Planning.</li> </ul>	The study was undertaken in a Paediatric Intensive Care Unit in a UK tertiary referral centre.

					<ul style="list-style-type: none"><li>• Potential benefits of Advance Care Planning include providing the opportunity to make decisions regarding end-of-life care in a timely fashion.</li><li>• Barriers to the process include the recognition of the life-limiting nature of an illness and gaining consensus of medical opinion.</li></ul>	
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Author, Year and Country	Aim/objective	Study design	Sample	Data collection techniques	Key findings	Comments
1. Basu and Swil (2018) Australia	Assess physicians' experiences and education regarding advanced care planning in paediatrics.	Quantitative.	Paediatricians (generalists and subspecialists) and paediatric advanced trainees were invited to participate. Ninety-two responses were obtained over a 10-week period.	Survey comprising of 25 multiple-choice questions. The questionnaire was formulated after the literature review in consultation with a senior paediatric intensivist and paediatric palliative care physician.	<ul style="list-style-type: none"> <li>• Patients with life-limiting conditions are encountered frequently, with 57% of respondents caring for at least 10 such patients during the last 2 years.</li> <li>• In total, 64% of respondents felt that advanced care planning discussions should occur early around the time of diagnosis or during a period of stability; however, 57% observed discussions occurring late in illness after multiple acute, severe deteriorations.</li> <li>• In total, 46% felt that multidisciplinary teams were the most appropriate to initiate advanced care planning discussions.</li> <li>• Prognostic uncertainty was the most common barrier to advanced care planning initiation.</li> <li>• Lack of experience and education were</li> </ul>	Confined to one professional group. Pertinent findings regarding decision-making also revealed. Basic trainees were excluded given their lesser clinical experience on average and hence less experience of advanced care planning. The study is limited by a small sample size and by being conducted at a single hospital site, which may result in respondents having limited variety in their experience of how advanced care planning is performed.

					<p>identified as barriers by 43 and 32%, respectively.</p> <ul style="list-style-type: none"> <li>The majority of respondents regarded exposure to advanced care planning and education during training as inadequate.</li> </ul>	
2. Coad, Patel and Murray (2014) United Kingdom	Explore the problem of communication barriers in pediatric palliative care, particularly the detrimental effects of poor interaction between staff and families on children's health and well-being.	Literature review.	Databases were searched from 1996 to 2011 and included Academic Search Complete, ASSIA, CINAHL, Google Scholar, Medline, PsycInfo, and Scopus.	A total of 15 articles were included.	<p>Five themes were identified:</p> <ul style="list-style-type: none"> <li>Poor staff communication skills. Parents are not always satisfied with how staff members disclose diagnostic information.</li> <li>Problematic information exchange. A cause for concern is the wide variety of professionals who provide diagnostic information and their varying levels of clinical experience.</li> <li>Language barriers. Communication is difficult when the patient or family members do not speak the same language as health care staff.</li> <li>Complex care needs. Staff report that at times during the course of</li> </ul>	Eleven articles were identified from the results reporting on communication barriers within palliative care for children and young people and four others reported on communication barriers specifically during diagnostic disclosure.

					<p>providing clinical feedback to children and families, despite their best efforts, conflicts arise.</p> <ul style="list-style-type: none"> <li>• Training and education needs. There is limited educational preparation regarding communication.</li> </ul>	
3. Hill et al. (2013) United States	<p>1. Describe the problems and hopes reported by mothers, fathers and physicians of children receiving palliative care.</p> <p>2. Examine the observed concordance between participants.</p> <p>3. Examine parental perceived agreement.</p> <p>4. Examine whether parents who identified specific problems also specified corresponding hopes, or whether the problems were left 'hopeless'.</p>	Mixed method.	<p>71 parents (44 mothers, 25 fathers, one stepmother and one grandmother ) of children with life-limiting conditions and 43 physicians. The stepmother and the grandmother were classified as mothers.</p>	<p>Semi-structured interviews. Parents and physicians were asked to report problems and hopes and perceived agreement for 50 children receiving palliative care. Problems and hopes were classified into eight domains. Observed concordance was calculated between parents and between each parent and the physicians.</p>	<ul style="list-style-type: none"> <li>• The most common problem domains were physical body (88%), quality of life (74%) and medical knowledge (48%).</li> <li>• The most common hope domains were quality of life (88%), suffering (76%) and physical body (39%).</li> <li>• Overall parental dyads demonstrated a high percentage of concordance (82%) regarding reported problem domains and a lower percentage of concordance on hopes (65%).</li> <li>• Concordance between parents and physicians regarding specific children was lower on problem (65–66%) and hope domains (59–63%).</li> <li>• Respondents who identified</li> </ul>	<p>The findings are also pertinent with regard to decision making.</p>

					problems regarding a child's quality of life or suffering were likely to also report corresponding hopes in these domains (93 and 82%, respectively).	
4. Hunter (2017) United Kingdom	Describe the nature of work within a children's hospice and in particular the role of a paediatrician working within this setting.	Review of the epidemiology of paediatric palliative care and current delivery of care. The author will describe the nature of work within a children's hospice and in particular the role of a paediatrician working within this setting.	One paediatrician	Review.	<p>The author concludes:</p> <ul style="list-style-type: none"> <li>• I continue to feel privileged to be able to work in the area of my choosing with a high degree of professional freedom.</li> <li>• I do have concerns about the paucity of NHS provision in this important area of paediatric medicine and believe that we need to do more to increase access to specialist medical input and to increase working across the boundaries between NHS and hospices.</li> <li>• Expectations of receiving expert medical care in all areas rightly continue to rise and it is difficult to see how hospices can continue to meet this expectation without partnerships with NHS providers that allow the</li> </ul>	One professional's perspective. Valuable information regarding experiences and service provision.

					<p>medical specialists to be properly supported.</p> <ul style="list-style-type: none"> <li>Equally NHS services can benefit greatly from the perspective and skills brought by those working within a hospice organisation.</li> </ul>	
5. Price, McCloskey and Brazil (2017) United Kingdom	To report parent and professional perspectives of step-down care in assisting the transition from hospital to home, within one children's hospice in a constituent country of the United Kingdom.	Qualitative.	5 parents (of 4 children) who had received step-down care in one specified children's hospice. 20 professionals were recruited on the basis they had experience of working with any of the 4 children and their families during the transition from hospital towards home.	Interviews with parents and focus groups with healthcare professionals.	<ul style="list-style-type: none"> <li>Multiple benefits of step-down in the hospice were clear.</li> <li>Both sets of accounts suggested that for children and families life was "on hold" in hospital.</li> <li>Hospice was considered a home-like environment where the child and family could "live again".</li> <li>Parents reflected that, in hospice they were "living, not existing" while professionals highlighted hospice as nurturing and empowering the whole family, promoting the child's development while safely meeting their clinical needs.</li> </ul>	All of the children had non-malignant life-limiting conditions.
6. Rodriguez and King (2014)	Explore the lived experience of caring and	Qualitative.	21 paediatric palliative care	Focus-groups with professionals	<ul style="list-style-type: none"> <li>Parents' expectations for support were</li> </ul>	The findings also revealed issues

United Kingdom	care planning for a child with a life-limiting condition.		professionals and 20 parents of children with life-limiting conditions.	and semi-structured interviews with parents.	<p>raised by the diagnosis, but the reality could disappoint, which put pressure on professionals.</p> <ul style="list-style-type: none"> <li>• Current service designs with respect to key working did not always coincide with family preference.</li> <li>• Both parents and professionals found that the care journey required them to shift personas to respond to different contexts.</li> </ul>	surrounding access to services.
7. Trowbridge et al. (2017) United States	Illustrate themes that emerge when providing palliative care for children with rare diseases: a case series of three children with congenital disorder of glycosylation.	Report.	Parents of 3 children with life-limiting conditions gave permission for their child's name and story to be shared.	Description and analysis of three cases.	<ul style="list-style-type: none"> <li>• Despite similar diagnoses and outcomes, these three cases highlighted unique challenges in the medical, psychosocial, and spiritual realms.</li> <li>• Three pertinent themes were discussed.</li> <li>• Decision making as a dynamic process-for each family, decision making was not a single act; each diagnostic test, intervention, or medication was delicately considered based on its potential to help versus potential to harm.</li> <li>• The challenge of providing</li> </ul>	The findings also revealed issues surrounding decision-making.

					<p>palliative care in the face of uncertainty-acknowledging the frustration of uncertainty helped families to cope and shift their focus from the unanswerable to the questions that had to be answered with the limited information at the time.</p> <ul style="list-style-type: none"> <li>• The necessity of the comprehensive psychosocial-spiritual assessment and intervention.</li> </ul>	
8. Monterosso et al. (2007) Australia	To obtain feedback from families of children receiving palliative and supportive care about their care needs in hospital and in community settings. To elicit the views of service providers.	Mixed method in two phases.	134 parents of children receiving palliative care and 20 service providers.	Phase 1 consisted of a series of surveys and Phase 2 consisted of interviews with parents and service providers.	<ul style="list-style-type: none"> <li>• The concept of palliative care is poorly understood by health professionals and by parents.</li> <li>• Parents indicated the need for clear and honest information about their child's condition and prognosis.</li> <li>• They perceived this had been lacking.</li> <li>• Families required financial and practical assistance with providing care.</li> <li>• The level of respite (in home and residential) was perceived to be insufficient and inequitable.</li> </ul>	Parents of children with cancer and non-malignant conditions were included. The parents in the cancer group were bereaved whereas in the non-malignant group they were not. The majority of the parents who participated were in the non-malignant group.

					<ul style="list-style-type: none"><li>• There was a perceived lack of coordination between community services and the hospital.</li></ul>	
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Author, Year and Country	Aim/objective	Study design	Sample	Data collection techniques	Key findings	Comments
1. Abode et al. (2011) United States	Explore barriers to hospital discharge for young children with new tracheostomies.	Chart review.	A subset of 20 (29%) children with length of stay (LOS) longer than the mean of 44 days underwent chart review to identify factors associated with prolonged LOS.	Demographics, hospital discharging unit, and discharge disposition were identified and inpatient narrative notes were scrutinized.	<ul style="list-style-type: none"> <li>• Overall mean age at tracheostomy was 6 months, versus 3.2 months for children with prolonged LOS.</li> <li>• For the overall group, 29% were discharged from the neonatal unit versus 60% of the prolonged LOS group.</li> <li>• Discharge to a longterm care facility occurred in 10.1% of the overall group versus 30% of the prolonged LOS group.</li> <li>• Contributing factors were represented individually or in combination for all 20 children with prolonged LOS.</li> </ul>	Presence of disease alone was associated with prolonged LOS for 8 (40%) children. Other barriers to discharge included unavailability of long term care beds or home nursing; inadequate family resources and supports; and delays in completion of home caregiver training

<p>2. Amin et al. (2015) Canada</p>	<p>Assess the length of stay required to initiate long-term invasive ventilation at the authors' institution, which would inform future interventional strategies to streamline the in-hospital stay for these families.</p>	<p>Chart review of children initiated on invasive long-term ventilation via tracheostomy between January 2005 and December 2013.</p>	<p>35 children initiated on long-term invasive ventilation.</p>	<p>Health care records were reviewed to obtain the following information: date of admission; reason for admission; date of tracheostomy insertion; diagnosis; discharge date from the acute care hospital; discharge destination; duration of stay at the rehabilitation centre; date of death (if applicable); location of death (if applicable); current location of residence. If the patient was never discharged home, the last date of the study was considered to be the 'discharge date' to calculate length of stay.</p>	<ul style="list-style-type: none"> <li>• 35 children were initiated on long-term invasive ventilation via tracheostomy at the acute care hospital.</li> <li>• Musculoskeletal disease was the most common reason for tracheostomy insertion.</li> <li>• 2 children died during the hospital admission.</li> <li>• 15 children were discharged home directly from the acute care hospital.</li> <li>• 18 were moved to the rehabilitation centre and were never discharged home.</li> <li>• Combining the length of stay at the acute care and rehabilitation centre for the entire cohort, the median length of stay was 162 days from the time of tracheostomy insertion.</li> </ul>	<p>The median length of stay from the initiation of invasive long-term ventilation to discharge home from the rehabilitation was somewhat long compared with other ventilation programs world wide. Additionally approximately 20% of the cohort never transitioned home.</p>
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3. Bona, Bates and Wolfe (2011) United States	To describe Massachusetts' experience in implementing a novel pediatric palliative care program.	Qualitative.	Families of 227 children with life-limiting illness enrolled in a paediatric palliative care program.	Written responses to a family satisfaction survey. Enrollment and service trends were identified using Massachusetts Department of Public Health administrative data.	<ul style="list-style-type: none"> <li>• There were 11 deaths, 100% of which occurred in the family's requested location.</li> <li>• Median length of stay on service prior to death was 233 days.</li> <li>• Families most commonly implemented psychosocial and case management services, followed by complementary therapies, and volunteer services.</li> </ul>	Admitting diagnoses included cancer (30%), chromosomal abnormalities (17%), neurodegenerative disorders (15%), and other (38%). The authors concluded that successful implementation of a statewide pediatric palliative care program as modeled in Massachusetts is highly feasible at relatively low cost.
4. Boyden et al. (2018) United States	Explore factors that are associated with the use of community-based pediatric palliative care for U.S. children with life-limiting illnesses and life-threatening illnesses and their families.	Integrative review.	A literature search of PubMed, CINAHL, Scopus, Google Scholar, and an ancestry search was performed to identify empirical studies and program evaluations published between 2000 and 2016.	40 peer-reviewed quantitative and qualitative methodological interdisciplinary articles were included in the final sample.	<ul style="list-style-type: none"> <li>• Patient characteristics such as older age and a solid tumor cancer diagnosis and interpersonal factors such as family support were associated with community-based pediatric palliative care higher use.</li> <li>• Organizational features were the most frequently discussed factors that increased community-based pediatric</li> </ul>	Multilevel factors are associated with increased community-based pediatric palliative care use for children with life-limiting or life-threatening illnesses and their families in the United States.

					<p>palliative care, including the importance of interprofessional hospice services and interorganizational care coordination for supporting the child and family at home.</p> <ul style="list-style-type: none"> <li>• Finally, geography, concurrent care and hospice eligibility regulations, and funding and reimbursement mechanisms were associated with community-based pediatric palliative care use on a community and systemic level.</li> </ul>	
5. Bradford et al. (2012) Australia	Identify barriers and gaps and to plan for the future of the Paediatric Palliative Care Service of the Royal Children's Hospital in Brisbane.	Qualitative.	Stakeholders, including medical staff specialists (24%), nurses (32%), allied health (24%), community support group representatives (16%), corporate policy representatives	Focus groups.	<ul style="list-style-type: none"> <li>• A number of barriers were highlighted including: equity in access to services; awareness, understanding and fear; experience of health professionals; funding and resources.</li> </ul>	The 13 focus groups consisted of four to five individuals and were pre-organised to mix disciplines as well as community and hospital representatives, encouraging networking and provoking thought and discussion beyond individuals'

			<p>ves (2%) and bereaved parents (2%) from South East Queensland (SEQ) were invited to attend a forum.</p>		<ul style="list-style-type: none"> <li>• A lack of respite options, the need for further education, improved collaboration and improved communication between services were identified as existing gaps</li> </ul>	<p>usual areas of expertise</p>
<p>6. Doug et al. (2011) United Kingdom</p>	<p>To evaluate the evidence on the transition process from child to adult services for young people with palliative care needs.</p>	<p>Systematic review.</p>	<p>The bibliographic databases Medline, CINAHL, PsychINFO, British Education Index and the International Bibliography of the Social Sciences from 1995 to 2008. The authors also searched the reference lists of included studies and sought additional literature through contact with experts in palliative care.</p>	<p>92 papers met the inclusion criteria and were from a variety of countries around the world.</p>	<ul style="list-style-type: none"> <li>• There is no evidence of transition services that address 'palliative care' care as an overall concept.</li> <li>• Transition programs that exist are mainly condition-specific.</li> <li>• No long-term outcome data exist to compare the effectiveness/cost effectiveness of the condition-specific or generic transition models.</li> <li>• Some consensus exists on what may facilitate or impede successful transition programs.</li> </ul>	<p>Studies were included if participants were as follows: (1) Young people aged 13-24 years with palliative care conditions; (2) relevant to situational transition, that is from child services to adult services and/or between healthcare providers and (3) original articles, reviews, systematic reviews consensus policies, guidelines and survey recommendations.</p>

					<ul style="list-style-type: none"> <li>• Transition should be multidisciplinary in nature, individualised in terms of addressing need and navigated with the help of a designated key worker.</li> </ul>	
7. Hunt et al. (2015) United Kingdom	Gain a more informed picture of issues affecting children/young people with life limiting conditions, their parents and local professionals prior to preparing a bid to evaluate services.	Consultation with service users and providers.	Two families and ten health-care professionals participated, including paediatricians, a children's hospice representative and representatives from three community children's nursing teams. Six children aged from 13-18years.	Multiple methods included focus groups, face-to-face interviews and telephone interviews.	<ul style="list-style-type: none"> <li>• An overarching theme from parents was 'Why does it happen like this?' Services did not seem designed to meet their needs.</li> <li>• Whilst children and young people expressed ideas related to quality of environment, services and social life, professionals focused on ways of meeting the families' needs.</li> <li>• The theme that linked families' concerns with those of professionals was 'assessing individual needs'.</li> </ul>	Challenges regarding recruitment were highlighted. Only two families were recruited.

<p>8. Kerr et al. (2017) United Kingdom</p>	<p>To explain how intervention processes interact with contextual factors to help transition from children's to adult services for young adults with life-limiting conditions.</p>	<p>Systematic realist review of the literature.</p>	<p>Literature was sourced from four electronic databases: Embase, MEDLINE, Science Direct and Cochrane Library from January 1995 to April 2016. This was supplemented with a search in Google Scholar and articles sourced from reference lists of included papers.</p>	<p>78 articles were included in the review.</p>	<ul style="list-style-type: none"> <li>• Six interventions were identified related to an effective transition to adult services.</li> <li>• Contextual factors include the need for children's service providers to collaborate with adult service providers to prepare an environment with knowledgeable staff and adequate resources.</li> <li>• Mechanisms triggered by the interventions include a sense of empowerment and agency amongst all stakeholders.</li> </ul>	<p>Data were extracted using an adapted standardised data extraction tool which included identifying information related to interventions, mechanisms, contextual influences and outcomes. Two reviewers assessed the relevance of papers based on the inclusion criteria.</p>
<p>9. Kirk and Glendinning (2004) United Kingdom</p>	<p>To explore the experiences of families caring at home for a technology-dependent child; to examine their needs for practical and other support; and to examine how far services are currently</p>	<p>Qualitative.</p>	<p>Parents of 24 technology-dependent children and with 44 healthcare, social care and other professionals</p>	<p>In-depth interviews.</p>	<ul style="list-style-type: none"> <li>• Services in the community were not sufficiently developed to support this group of families.</li> <li>• Major problems were identified in the purchasing and provision</li> </ul>	<p>The authors state that services need to work in partnership with families and with each other at both strategic and operational levels, to develop integrated and coordinated services that can meet the needs of this group of families.</p>

	meeting these needs.				<p>of short term care/home support services and specialist equipment/therapies in the community.</p> <ul style="list-style-type: none"> <li>• Service provision could be poorly planned and coordinated at an operational level and few families had a designated key worker.</li> <li>• Parents felt that professionals did not always recognise either the emotional costs entailed in providing care of this nature or their expertise in caregiving.</li> <li>• Information-giving to parents was often described as poor and participants reported that hospital professionals failed to negotiate the transfer of caregiving responsibility to parents.</li> </ul>	It was unclear as to how many children had life-limiting conditions.
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<p>10. Liberman et al. (2016) United States</p>	<p>This study evaluated whether introducing palliative care during primary care appointments (1) was feasible; (2) increased access and improved knowledge of palliative care; and (3) facilitated advanced care planning.</p>	<p>Pilot study of a multi-modal intervention including targeted education for primary care providers, an information packet for families and presence of a palliative care team member in the outpatient clinic.</p>	<p>Primary care providers and families of children with complex chronic medical conditions.</p>	<p>Primary care providers completed pre- and post-surveys assessing experience, knowledge and comfort with palliative care. Enrolled families received an information packet; a subset also met a palliative care team member. All families were encouraged to make an appointment with the palliative care team, during which the team assessed palliative care needs and goals of care. Upon study completion, the investigators assessed family and primary care provider satisfaction and collected feedback on project feasibility.</p>	<ul style="list-style-type: none"> <li>• 20 families were enrolled and received the information; 15 met a palliative care team member.</li> <li>• Of the 17 families who completed a post-study survey, 11 had never heard of palliative care and 13 were unaware that the team existed.</li> <li>• Most families perceived palliative care information as 'very important'.</li> <li>• All would recommend palliative care services to others.</li> <li>• 9 families followed up with the team, but were not prepared to complete an advanced care plan.</li> <li>• Providers reported lack of training in communicating bad news and conducting goals of care discussions.</li> </ul>	<p>The authors concluded that initiating palliative care services in the outpatient primary care setting is logistically challenging but increases access to palliative care for children with complex chronic medical conditions and improves palliative care knowledge and comfort for primary care providers. They felt increasingly comfortable introducing palliative care to families and supported program continuation.</p>
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<p>11. Pentaris (2018) United Kingdom</p>	<p>Explore palliative care professional's perceptions of barriers and challenges to accessing children's hospice and palliative care services.</p>	<p>Qualitative.</p>	<p>5 palliative care professionals , working across tertiary and community-based services.</p>	<p>Individual semi-structured interviews.</p>	<p>Three themes emerged from the data.</p> <ul style="list-style-type: none"> <li>• The first theme refers to the current availability and adequacy of palliative and hospice care in addressing the children's and families' needs.</li> <li>• The second theme is concerned with the main barriers that interfere with or prevent the use of the services.</li> <li>• The third theme is concerned with the role of cultural and family values/ priorities in accessing children hospice and palliative care.</li> </ul>	<p>The palliative care professionals were working in nursing or social work roles. Three were involved in making referrals to children's hospice services (two worked in a children's hospital and one in a charity for children with life-limiting or threatening conditions) and the other two worked in a children's hospice.</p>
<p>12. Vollenbroich et al. (2012) Germany</p>	<p>Evaluate the effectiveness of a specialized pediatric palliative home care team as experienced by parents and healthcare professionals.</p>	<p>Quantitative.</p>	<p>Parents and healthcare professionals involved in the care of terminally ill children who died and whom the pediatric palliative home care team was in charge of.</p>	<p>Parental questionnaire 1 comprised a total of 56 items and was developed specifically for this study. Parental questionnaire 2 comprised validated questionnaires to assess anxiety and depression.</p>	<ul style="list-style-type: none"> <li>• Satisfaction with the pediatric palliative home care team scored a median of 10 (numeric rating scale, 0-10).</li> <li>• The child's death was perceived as predominantly peaceful (median, 9);</li> </ul>	<p>Congenital (including congenital heart disease) and oncologic diseases were the predominant underlying disorders. A number of children also died as a result of neurological conditions.</p>

			<p>43 parent dyads participated.</p> <p>87 healthcare professionals participated, including physicians, nurses, social workers, psychologists and case managers.</p>	<p>The questionnaire for the healthcare professionals was developed specifically for this study.</p>	<p>71% died at home.</p> <ul style="list-style-type: none"> <li>• According to parents, involvement of the pediatric palliative home care team led to highly significant (<math>p &lt; 0.001</math>) improvements in the children's symptoms and quality of life, as well as in aspects of communication and administrative barrier reduction.</li> <li>• Anxiety was detected in 25% of parents, depression in 19%, and prolonged grief disorder in 13%.</li> <li>• Healthcare professionals evaluated all investigated care domains as being significantly improved (<math>p &lt; 0.001</math>).</li> <li>• 35% of healthcare professionals felt uncertain concerning pediatric palliative care, 79% would</li> </ul>	
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					welcome specific training opportunities.	
13. Whitton et al. (2008) United Kingdom	To evaluate a new rural community palliative care service for children according to the perceptions of families and service providers, to make changes suggested by families and to re-evaluate 1 year later.	Evaluation.	24 families completed postal questionnaires in 2005. 27 families completed postal questionnaires in 2006. Two families also participated in interviews in 2006. 12 healthcare professionals participated in 2005 and 15 in 2006.	In 2005, 2 years after the onset of the service, families were sent postal questionnaires, including the Measure of Process of Care (MPOC-UK). In 2006, all of the families receiving care from the service were given the option of completing the questionnaire. In both years, the service providers were asked to complete the Measure of Process of Care for Service Providers (MPOC-SP). Two families also completed qualitative interviews about their experience of the service.	<ul style="list-style-type: none"> <li>• Seven (29%) of families completed the survey in 2005.</li> <li>• Families rated 'Respectful and Supportive Care' as the highest domain in the MPOC-UK and 'Providing General Information' as the lowest.</li> <li>• Particular emphasis was placed on improving provision of information during the following year.</li> <li>• Fourteen (52%) families completed the survey in 2006.</li> <li>• Scores increased across all domains in the second survey.</li> <li>• The largest increase was 'Providing General Information'.</li> </ul>	The results from both of the MPOC tools were extremely useful in helping providers to identify aspects of the service in need of improvement and hence implement valued changes.

Author, Year and Country	Aim/objective	Study design	Sample	Data collection techniques	Key findings	Comments
1. Atout, Hemingway and Seymour (2017) Jordan	Explore the experience of decision making in the care of children with palliative care needs in Jordan, from the perspective of their mothers.	Collective qualitative case study approach.	Data were collected in 3 pediatric wards in a Jordanian hospital. 24 mothers, 12 physicians and 20 nurses participated.	Participant observation (197 observational hours) and 56 semi-structured interviews with the mothers, physicians and nurses.	<ul style="list-style-type: none"> <li>The findings show how Jordanian mothers seek to transfer the role of decision making to physicians, as they perceive themselves to be unable to make decisions about critical issues related to the treatment of their children.</li> <li>Mothers had a widespread apprehension of "future guilt," especially when they feared that any decisions they might make could have an adverse impact on their children.</li> <li>Contrary to the predominant pattern, some mothers took a proactive approach towards decision making about their children's treatment.</li> </ul>	The authors conclude that mothers prefer to involve physicians in decisions about their children's healthcare and treatment to eliminate their fear of probable future guilt; this modifies any tendency to autonomously decide for their children. These findings are underpinned by the Jordanian culture in which doctors' opinions are highly regarded.

					<ul style="list-style-type: none"> <li>• These mothers requested detailed information from primary physicians and sought different sources of knowledge such as second opinions, reading online resources, or talking to other parents.</li> </ul>	
2. de Vos et al. (2015) Netherlands	Explore how physicians and parents communicate about end-of-life decisions and to what extent parents share in the decision-making process.	Prospective exploratory study.	Overall, 27 physicians participated, along with 37 parents of 19 children from whom a decision to withhold or withdraw life-sustaining treatment was being considered.	Forty-seven conversations were audio-recorded, ranging from 1 to 8 meetings per patient.	<ul style="list-style-type: none"> <li>• All physicians focused primarily on providing medical information, explaining their preferred course of action, and informing parents about the decision being reached by the team.</li> <li>• Only in 2 cases were parents asked to share in decision-making.</li> <li>• Despite their intense emotions, most parents made great effort to actively participate in the conversation.</li> <li>• They did this by asking for clarifications,</li> </ul>	The study was conducted in the Paediatric Intensive Care Units (PICU) of 2 Dutch university medical centers. Both PICUs are combined medical and surgical tertiary facilities, including all specialty care except that involving prematurely born infants. A large variation of cases according to the following patient-related characteristics: age, gender, prognosis, preexisting neurologic damage, total duration of care and length in PICU were included. It was unclear as to how many children had life-limiting conditions.

					<p>offering their preferences, and reacting to the decision being proposed (mostly by expressing their assent).</p> <ul style="list-style-type: none"> <li>• In the few cases where parents strongly preferred life-sustaining treatment to be continued, the physicians either gave parents more time or revised the decision.</li> </ul>	
<p>3. Dunlop (2008) United Kingdom</p>	<p>The dying child: should we tell the truth? A review of three perspectives: the child, the parents and the professionals.</p>	<p>Literature review.</p>	<p>Studies focusing on children with life-limiting or life-threatening conditions.</p>	<p>It was unclear as to how many papers were included in the review.</p>	<ul style="list-style-type: none"> <li>• A review of these three perspectives on truth telling reveals some of the many factors that influence openness, communication and truthfulness.</li> <li>• Creation of a truthful care environment requires development of appropriate skills and attitudes in professionals, use of research evidence, ongoing assessment of the child and family's</li> </ul>	<p>Multiple perspectives considered. Very limited information regarding the structure of the review.</p>

					communicatio n needs and support for members of the multidisciplina ry care team.	
4. Gillam and Sullivan (2011) Australia	Review the current evidence regarding decisions about treatment limitation for young children with life-threatening or life-limiting conditions with the aim of answering three questions. 1. What actually happens in practice-are parents involved in end-of-life decisions for their children? 2. Do parents want to be involved in making these decisions? 3. What is the effect on parents on having been involved?	Literature review.	Studies which incorporate end-of-life decision-making where a child has a life-limiting or life-threatening condition.	16 studies were included.	<ul style="list-style-type: none"> <li>• The literature suggests that parents do want to be involved in end-of life decision-making for their child.</li> <li>• The nature of the involvement they are seeking can vary, from being included in discussions to making and taking responsibility for the final decision.</li> <li>• Parents who believe that they have been involved and even that they have taken final responsibility for a decision that has resulted in the death of their child are mostly not negatively affected by this.</li> <li>• Feelings of guilt or regret are no more frequent for them than for</li> </ul>	No details were provided regarding how the literature review was undertaken.

					parents who have not been decision-makers.	
5. Heckford and Beringer (2014) United Kingdom	To review advance care planning for children with life-threatening or life-limiting conditions.	Retrospective case note review.	Study subjects were from two National Health Service Trusts in Bristol in the United Kingdom. Cases were identified from Child Death Overview Panel data.	Forty-two sets of case notes were reviewed in relation to 20 children. Measurements quantitative and qualitative review of advanced care planning in relation to standards set by the Association of Children's Palliative Care (ACT).	<ul style="list-style-type: none"> <li>• In 25% of cases there was no documented discussion with families about the approach to end of life.</li> <li>• In 25% of cases there was no evidence of an advance care plan, and the content and accessibility of those that did exist was variable.</li> <li>• Forty-five percent of families were not offered a choice with regard to location of care in the last months of life and 50% were not offered a choice about location of death.</li> </ul>	Fifteen children had non-malignant conditions. The conditions included metabolic, complications of prematurity, neurological, cardiac and syndromic/ chromosomal. Five of the children had oncological conditions.
6. Knapp et al. (2009) United States	Validate the psychometric properties of the Decisional Conflict Scale (DCS) and the COMRADE instruments for children with life-limiting illnesses.	Quantitative.	266 parents whose children have life-limiting illnesses.	Analyses were conducted using survey data collected from the parents whose Medicaid enrolled children have life limiting illnesses.	<ul style="list-style-type: none"> <li>• Results of the analyses suggest that the DCS has better psychometric properties for measuring decision making within the population of children with life-</li> </ul>	Several study limitations merit attention. First, the response rate for the survey was 54.4%. Although this response is consistent with other surveys conducted with Medicaid eligible populations, there

					<p>limiting illnesses than the COMRADE.</p>	<p>may be inherent differences between responders and non-responders. Second, Medicaid eligible children tend to have high utilization of healthcare services and lower health outcomes, both of which could affect their parents' decision-making experiences.</p>
<p>7. Michelson et al. (2013) United States</p>	<p>Describe the roles and respective responsibilities of pediatric intensive care unit (PICU) health care professionals (HCPs) in end-of-life care decisions faced by PICU parents.</p>	<p>Retrospective qualitative study.</p>	<p>Eighteen parents of children who died in the pediatric ICU and 48 PICU healthcare professionals (physicians, nurses, social workers, chaplains, and case managers).</p>	<p>In depth, semi-structured focus groups and one-on-one interviews.</p>	<ul style="list-style-type: none"> <li>• The family supporter addresses emotional, spiritual, environmental and informational needs.</li> <li>• The family advocate helps families articulate their views and needs.</li> <li>• The information giver provides parents with medical information.</li> <li>• The general care coordinator helps facilitate interactions among healthcare professionals and between healthcare professionals and parents.</li> <li>• The decision maker makes</li> </ul>	<p>The authors outline that while HCPs described all end-of-life care decisions they perceive that parents face, the majority of their discussion focused on decisions related to limitation of therapies.</p>

					<p>or directly influences the defined plan of action.</p> <ul style="list-style-type: none"> <li>• The end-of-life care coordinator organizes and executes functions occurring directly before, during, and after dying/death.</li> <li>• The point person develops a unique trusting relationship with parents.</li> </ul>	
8. Peay et al. (2016) United States	Explore clinicians' perspectives and parents' decision making about children's participation in Duchenne muscular dystrophy (DMD) clinical trials.	Qualitative.	Eleven clinicians involved in ten trials and fifteen parents involved in six trials.	Semi-structured interviews.	<ul style="list-style-type: none"> <li>• Parents described benefit-risk assessments using information from advocacy, peers, professionals, and sponsors.</li> <li>• Strong influence was attributed to the progressive nature of DMD.</li> <li>• Most expected direct benefit.</li> <li>• Few considered the possibility of trial failure.</li> <li>• Most made decisions to participate before the</li> </ul>	The primary limitation of the study is that it was retrospective in that parents were asked to think back to their decision-making process. The timing of the deliberation and informed consent varied; for some parents that process occurred relatively close to the date of the interview, while for others it occurred several years in the past. Once a determination to participate is made, it is possible that parents re-frame their perceptions to be consistent with their decision.

					<p>informed consent process, but none-the-less perceived informed choice with little to lose for potential gain.</p> <ul style="list-style-type: none"> <li>• Clinicians described more influence on parental decisions than attributed by parents.</li> <li>• Clinicians felt responsible to facilitate informed consent while maintaining hope.</li> <li>• Both clinicians and parents reported criticisms about the informed consent process and regulatory barriers.</li> </ul>	
9. Popejoy et al. (2017) United Kingdom	To synthesize findings from qualitative research that has investigated decision-making and future planning for children with life-limiting conditions.	A qualitative systematic review and thematic analysis.	The bibliographic databases CINAHL, Medline, Embase and PSYCHInfo were searched. In order to find any relevant literature outside of the health field, a free	887 papers were identified for review and 5 papers were selected for inclusion, using predefined criteria.	<ul style="list-style-type: none"> <li>• Nineteen descriptive themes were identified, and these were further synthesized into four analytical themes.</li> <li>• Analytical themes were 'decision factors', 'family factors',</li> </ul>	Included papers focused on a range of stakeholders. The included studies collectively had a sample of 112 parents or guardians, 35 children and 14 professionals.

			text search was conducted in ProQuest. Additionally, EThOS (E-Theses Online Service) was searched.		<p>‘relational factors’ and ‘system factors’.</p> <ul style="list-style-type: none"> <li>The synthesis identified that decision-making and future planning need to be individualized for the family and their circumstances.</li> </ul>	
10. Postier, Catrine and Remke (2018) United States	Highlight the ways in which the involvement of pediatric palliative care (PPC) teams can help to make the option for home compassionate extubation (HCE) available when families choose to spend their child’s end of life at home.	Case presentations.	Case presentaiions incorporatin g a fifteen year old girl and an eighteen month old boy.	2 case presentations.	<ul style="list-style-type: none"> <li>Though several challenges to realizing the option of HCE exist, early consultation with a PPC team in the hospital, the development of strategic community partnerships, early referral to home based care resources, and timely discussion of family preferences may help to make this option a realistic one for more families.</li> </ul>	The cases presented demonstrate how families’ wishes with respect to how and where their child dies can be offered, even in the face of challenges.

<p>11. Xafis, Wilkinson and Sullivan (2015) Australia</p>	<p>Provide insight and guidance in the development of written resources (short print and online comprehensive version) for parents facing end-of-life decisions for their child.</p>	<p>A meta-analysis of predominantly empirical research pertaining to features which assist or impede parental end-of-life decisions.</p>	<p>The bibliographic database Scopus was used. Scopus uses a number of resources such as MEDLINE, EMBASE, open access papers as well as grey literature.</p>	<p>58 papers were included.</p>	<ul style="list-style-type: none"> <li>• The most prominently cited needs in the literature related to numerous aspects of information provision; the quantity, quality, delivery, and timing of information and its provision impacted not only on parents' ability to make end-of-life decisions but also on their emotional wellbeing.</li> <li>• The meta-analysis supports the value of written materials, as these provide guidance for both parents and healthcare professionals in pertinent content areas.</li> </ul>	<p>Included studies relating to critically ill children and newborn infants published between 1990 and 2012. It was unclear as to how many papers related to children with non-malignant life-limiting conditions. The search term palliative care was included.</p>
<p>12. Xafis et al. (2015) Australia</p>	<p>Discuss the development of the Caring Decisions handbook and website, modification after reviewer feedback and findings from initial pilot implementation</p>	<p>A targeted literature review; development phase-guided by the literature and the researchers' expertise; consultation process-</p>	<p>12 experts and parents formed the consultation panel. 8 parents of children with life-limiting conditions and clinicians were</p>	<p>Literature review, consultation and Interviews.</p>	<ul style="list-style-type: none"> <li>• Numerous factors supporting/impeding end of life decisions were identified.</li> <li>• Caring Decisions addressed issues identified in</li> </ul>	<p>The low level of dissemination of 'Caring Decisions' to parents during the study was a limitation of the pilot evaluation.</p>

		<p>comprised a multi-disciplinary panel of experts and parents; pilot evaluation study-hard-copy handbook was distributed as part of routine care.</p>	<p>interviewed in the pilot study.</p>		<p>the literature and by the multidisciplinary research team.</p> <ul style="list-style-type: none"> <li>• The consultation panel provided overwhelmingly positive feedback.</li> <li>• Pilot study parents found the resources helpful and comforting.</li> <li>• Most clinicians viewed the resources as very beneficial to parents and identified them as ideal for training purposes.</li> </ul>	
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