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PhD in Medicine

**Exploring Service Use and Preferences of Care
for Children with Life-Limiting
Neurodevelopmental Conditions and their
Families: A Mixed Methods Study**

**A thesis submitted to the University of Dublin, Trinity College, for the
Degree of Doctorate of Philosophy**

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2015

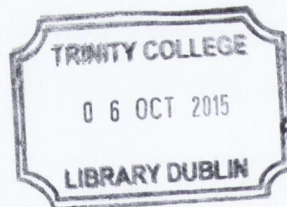
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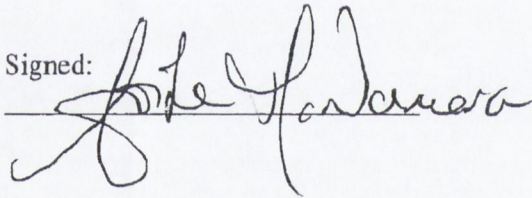


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Summary

A mixed methodological approach facilitated an in-depth examination of the service experiences, service preferences and financial costs associated with caring for children with life-limiting neurodevelopmental conditions at home in Ireland. A qualitative study using Interpretative Phenomenological Analysis (IPA) examined parents' lived experiences of caring for and using services in relation to their child. The IPA findings revealed the overwhelming nature of parents' experiences and needs around 8 major themes: (1) transitioning from hospital to home, (2) the perpetual wait for services, (3) the crucial role of liaison nurse (4) parents as advocates, (5) physiotherapy – a highly valued service (6) the financial costs of caring, (7) emotional effects of caring and (8) future concerns. A quantitative study using a Discrete Choice Experiment (DCE) elicited parents' preferences for hypothetically designed monthly care packages and revealed parents' valuation of services in the form of willingness to pay and willingness to trade for various services. Analysis revealed that parents have a preference for more of the following services for their child: physiotherapy, speech & language therapy, day nursing care and a cash maintenance payment. Finally, a descriptive study outlined the financial out of pocket costs associated with caring for a child with a life-limiting disability. Findings revealed that parents face significant out of pocket payments in relation to providing care to their child and lose on average up to fifty-percent of their household income. Having developed an evidence base and characterization of the range and scope of the issues facing families caring for children with life-limiting conditions, we provide recommendations to the Irish health care system.

Background note

This work was funded by the Health Research Board in Ireland under Grant No. PhD/2007/16. Aoife McNamara was a student on the Health Research board (HRB) Structured Population and Health Services Research Education programme (SPHeRE). This is a four year structured PhD programme with taught modules in year 1 and a research component conducted throughout the duration of years 2-4. The aim of the programme is to improve health through quality healthcare delivery and management in the Irish health care system. It is proposed that this will be achieved through the development of a critical mass of HSR expertise through PhD scholars who excel in research, policy evaluation and translation of evidence into practice. A more detailed overview of the HRB SPHeRE programme is provided in Appendix 1.

Dedication

**To
Alice Foy,
Eileen McNamara
&
Padraigh McNamara
(RIP)**

Acknowledgements

First and foremost, I would like to sincerely thank my supervisory team. I will always be grateful to Prof. Charles Normand for his continuous guidance, sage advice, and constant enthusiasm for this important topic. I am deeply indebted to have had the opportunity to work with him and am grateful for the superb learning experience. Sincere thanks also to Dr. Honor Nicholl who provided excellent guidance and feedback along the way and for encouraging me to present my work at various conferences.

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Finally and most of all, I would like to thank my husband Mark for his wonderful, kind, caring and patient support throughout this journey. The many conversations we shared in relation to this work inspired and encouraged me greatly throughout.

Table of Abbreviations

LLC's	Life-Limiting Conditions
ACT	Association for Children with Life-Limiting or Terminal Conditions and their Families
DOH&C	Department of Health and Children
CSO	Central Statistics Office
JJF	Jack & Jill Foundation
HSE	Health Service Executive
ICD	International Classification of Disease
UK	United Kingdom
CA	Canada
USA	United States of America
IHF	Irish Hospice Foundation
IPA	Interpretative Phenomenological Analysis
NIDD	National Intellectual Disability Database
NACPC	National Advisory Committee on Palliative Care
NDA	National Disability Authority
DCE	Discrete Choice Experiment
WTT	Willingness to Trade
WTP	Willingness to Pay
QALY	Quality Adjusted Life Year
SP	Stated Preference

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Chapter 1

Introduction to the Study

1.1 Introduction

Advances in medical and nursing care neonatal technology has led to a significant increase in the survival rates of children who are born with life-limiting conditions (Olsen and Maslin-Prothero, 2001). The management of chronic health conditions in children is extremely complex and requires support from a broad multidisciplinary team over the child's lifetime (Goldman, 1998). Children with life-limiting illnesses require life-sustaining care and paediatric palliative care interchangeably, and sometimes over decade long care trajectories (Noyes et al., 2013). There is a lack of evidence on how well Ireland's health care system meets the needs of these children in the community, although some Irish studies have identified areas of unmet need (Redmond and Richardson, 2003, Nicholl and Begley, 2012). Parents caring for a child with a life-limiting condition are in a unique position to assess how well their child's needs are being met. As they are often the primary carers for their child, parents typically have extensive knowledge on managing their child's illness. Furthermore, they have direct and in-depth experience in negotiating, accessing and delivering care to their child. Given all of the above, it is essential to gather and examine parents' perspectives to better understand the effective and ineffective components of the care provided to these children. Consequently, this study explores parents' experiences of, and preferences for, care services for children with life-limiting conditions living at home. A significant part of parents' experiences is the financial implications of caring, which is further explored in this study. The results of this study were obtained through interviews, administration of surveys and discrete choice experiments with families.

The purpose of this chapter is to provide an introduction to, and set the context for, the present study. It begins with a description of the background of the study and a brief description of the purpose of the study. The terms and definitions

used throughout are described, along with an overview of disability statistics; prevalence rates and mortality rates in relation to children with life-limiting conditions. The Irish care context is described and a discussion on Ireland's palliative care policy follows. The chapter concludes with a general overview of the study and an outline of the format of the remainder of the thesis.

1.2 Background

Due to technological advances in recent years, children born with complex disabilities are surviving longer than before. As a consequence therefore, the numbers of children needing care is rising. Rising life expectancy, coupled with advances in technology, increases the demand for care services in the community because much more can be done and is expected to be done for these children today (Craft, 2004). In order to evaluate and improve our current care models, an evidence base is needed. This will further support caregivers and health care providers in the development of services to meet the diverse needs of families across communities (Liben et al., 2008). In Ireland, it is estimated that the current number of children with life-limiting illnesses needing care is approximately 1,400 (Laura Lynn, 2013). These children represent a relatively small population in the Irish context but recent evidence shows that providing care to this specialised group is costly (Revill et al., 2013). The Irish government has acknowledged that more support and investment is needed for the provision of care at home (DOHC 2005) but, to date, little or no resources have been specifically earmarked for the provision of care to children with LLC's. This is the background that informs the rationale for this study.

1.3 Purpose of this Study

The purpose of this study is to provide a detailed evidence base for the development of care services that relate to children with life-limiting neurodevelopmental conditions at home in Ireland. The study progresses in two phases, the first of which examines parents' lived experiences of finding, accessing and providing care services for their child. In addition, the emotional support needs and financial challenges of providing care are explored. The phenomenon of parents' experience

is described, analysed and explicated in order to generate a greater understanding of parents' needs and expectations.

The second phase of this study involves a quantitative technique, where families participate in a quantitative questionnaire that aims to identify parents' preferences for hypothetically designed care packages. The technique quantifies parents' willingness to trade between services, which in turn, allows us to define a package of care to better meet the needs of this population group. Data was collected from parents across the Republic of Ireland using semi-structured in-depth interviews (n=24) and from subsequent quantitative questionnaires (n=69). A more detailed overview of research objectives are presented in chapter 3.

1.4 Definition of Key Terms

The next section outlines the terms and definitions used throughout the thesis to provide clarity and context for this study.

1.4.1 Definition of a Child

In Ireland, a 'child' is defined as a person under the age of 18 years, excluding a person who is or has been married (Department of Children and Youth Affairs, 2011). This is the accepted term used throughout the research literature.

1.4.2 Definition of Palliative Care

Children's palliative care is concerned with the treatment of children with life-limiting or life-threatening conditions. It is the need to maintain quality of life, not just in the dying stages, but also in the weeks, months and years before death and is characterized by a concern for symptom relief, promotion of general well-being and psychological and social comfort for the child and family (Craft, 2007). Adopted from Together for Short Lives, the definition of children's palliative care is as follows:

Palliative care for children and young people with life-limiting conditions is an active and total approach to care, from the point of diagnosis or recognition,

embracing physical, emotional, spiritual elements through to death and beyond. It focuses on enhancement of quality of life for the child/young person and support for the family includes the management of distressing symptoms, provision of short breaks and care through death and bereavement (Childrens Palliative Care Defintions, 2014).

Children's palliative care is a relatively new specialism and is distinct from adult's palliative care in that children's palliative care is often required over longer periods of time. Other factors to consider, is that conditions, often times, are extremely rare and children have unpredictable disease trajectories and prognoses. In contrast, adults who are receiving palliative care are typically diagnosed with cancer, have shorter periods of illness and have a predictable disease trajectory and prognosis. These distinctions highlight the need for specialist palliative care services and the necessity to identify the distinct needs of this population.

1.4.3 Changes in Defining Disability

Over the last decade, positive changes have occurred in the way disability is defined, which will have strong implications for the definitions, measurement and policy formulations for health and disability (WHO 2013). The World Health Organization (WHO) 1980's International Classification of Impairments, Disabilities and Handicap's (ICIDH) (WHO 1980) was redefined in 2004 to encompass a more positive attitude about disability. The original ICIDH model was criticized for its negative portrayal of the consequences of the disease in terms of disability and handicap. The updated International Classification of Function, Health & Disability (ICF) 2001 (WHO 2001) replaced the terms "impairment" and "handicap" with "body function and structure" and "activity". This represents a major transformation from emphasizing people's disability to focusing on their level of health. The ICF provides definitions, classifications and codes for its major components: body functions and structures, activities and participation, and environmental factors (Madden et al., 2012).

Moving from a medically focused model to a bio-psychosocial model, where contextual factors are considered, such as environmental factors (physical, cultural and social) and personal factors (gender, age, educational and lifestyle), forces

clinicians and care providers to think of the broader perspective when providing care. The ICF framework has the potential for changing the way clinicians assess care needs, as examined by Rosenbaum and Stewart (2004) in the context of children with Cerebral Palsy. Incorporating personal factors allows patients to challenge the traditional assumptions of what a “normal” level of functioning is, thus, allowing individuals to set their own personal standards of achievement in terms of what is more suitable for them. Rosenbaum and Stewart (2004) says the ICF model helps us to move away from a focus on “fixing” primary impairments to a view that places equal importance on encouraging functional activity and assisting the child’s involvement in all facets of life. The ICF framework is ideal for structuring patient problems, where the focus of intervention is not just placed on treatment of disease but on functioning and improvement of well-being (Martinuzzi et al., 2010)

1.4.4 Definition of a Life-Limiting Condition

Planning the provision of care services for children with life-limiting conditions is a difficult task due to a lack of consensus on the terms used; for example, in some cases, the term “life-limiting” is used, while in other cases, the term “life-threatening” is used, both of which require different care trajectories (Sutherland et al., 1992). This study uses the term “life-limiting” in the context of the definition outlined by the Association for Children with Life-Threatening or Terminal Conditions and their Families and the Royal College of Pediatrics and Child Health (2009) (ACT&RCP&CH). The definition is as follows: “Any illness in a child where there is no reasonable hope of cure from which the child or young adult will die”. A broad range of diagnoses lie under the remit of the term “life-limiting”, where some cases are rare and only encountered in the paediatric community, thus making paediatric expertise a necessity (Hynson and Sawyer, 2001). A common feature of children with life-limiting conditions is that they all have complex needs.

Ireland has adopted ACT& RCP&CH’s four defined categories of life-limiting conditions outlined in Table 1.1 below. The fourth category coincides with the type of illnesses children are diagnosed with in this study. Some diagnosis

examples include: Cerebral palsy, Rett Syndrome and Chromosomal deletion. For the purpose of this study, the term life-limiting condition will be used throughout, which encompasses a range of neurodevelopmental disabilities experienced by the children in this study.

Table 1.1 Four defined ACT categories

Group 1	Life-threatening condition for which curative treatment may be feasible but can fail. Palliative care may be necessary when treatment fails. Examples: Cancer, irreversible organ failures of heart, liver and kidneys.
Group 2	Conditions where premature death is inevitable, where there may be long periods of intensive treatment aimed at prolonging life & allowing participation in normal activities. Example: Cystic fibrosis.
Group 3	Progressive conditions without curative treatment options, where treatment is exclusively palliative and may commonly extend over many years. Examples: Batten’s disease, Mucopolysaccharidosis and Muscular Dystrophy
Group 4	Irreversible but non-progressive conditions causing severe disability leading to susceptibility to health complications and likelihood of premature death. Examples: Severe multiple disabilities, such as those following brain or spinal cord injuries, including some children with severe Cerebral Palsy

1.4.4.1 Definition of Neurodevelopmental Disability

Neurodevelopmental disability is a clinically distinct set of chronic disorders whose unifying feature is a documented disturbance in developmental progress, either quantitative or qualitative, or both, compared with established norms in one or more recognised developmental domains (American Psychiatric Association, 1994). Common features include; 1) an onset that is invariably during infancy or childhood, 2) an impairment or delay in the development of functions that are strongly related to biological maturation of the central nervous system and 3) a steady course that does not involve the remissions and relapses that tend to be

characteristic of other mental health disorders (Bishop and Rutter, 2008 p.50). Children with such disorders typically experience; motor (gross or fine), speech/language, cognition, personal/social, and challenges with activities of daily living (Shevell, 2009).

1.4.5 Complexity in Terminology

There is much ambiguity and inconsistency on what defines a life-limiting condition in the literature. In addition, frequent overlap in the provision of children's palliative care and disability care services exist (Laura Lynn, 2013). The lack of clear definitions presents many problems in the examination of prevalence rates, health care utilization patterns and service planning. Parents also face challenges, as a lack of definitions makes it difficult for them to identify service entitlements. The contrasting terminology used is widely acknowledged in the literature and has been raised as a serious issue in all three of Ireland's policy documents and reports relating to the planning of care services for children with LLC's (DOHC 2005, DOHC 2010, Laura Lynn, 2013). The most recent report by Laura Lynn (2013) emphasized the importance of promoting understanding of the terminology used among service providers and stakeholders. Research on terminology used by nurses in relation to children with LLC's in Ireland found that although there is a common term to use, the terminology used is often influenced by the diagnosis of the child (Nicholl, 2007). In Ireland, there is a myriad of terms used to describe children, which oscillates between 'life-threatening illnesses', 'complex care needs' and 'life-limiting illnesses'. The differences in terminology introduced complexity throughout the different phases of this study, and in particular when communicating with parents and nurses. A UK study by Lenton et al. (2001) found that three fundamental challenges exist when estimating the number of children with non-malignant LLC's: (1) an absence of clear definitions, (2) a wide range of diagnosis, with the probability of death being the only similarity between them, and (3) a lack of mutual services for all conditions.

1.5 Disability Statistics in Ireland & UK

In Ireland, the National Intellectual Disability Database (NIDD) reported a prevalence rate of (0.38/1000) children between the ages of 0-4 years having moderate to severe intellectual disability in 2012 (Kelly, 2013). The reported numbers are small but do however represent an increase over 2007's prevalence rate of (0.31/1000). The numbers may represent an underestimate of the true prevalence rates since 1) more children under the age of five have their needs met by conventional rather than specialized intellectual disability services 2) parents can be reluctant to provide information to a database on their child and 3) delays in diagnoses. All of these factors result in fewer children registering with the NIDD. According to the central statistics office (CSO) (Central Statistics Office, 2011) in 2011, 3.1% of children between the ages of 1 - 4 had a disability in Ireland. In the UK, 0.8 million children were reported as having some form of disability between the years 2011-12, which includes, longstanding illnesses, disabilities and infirmity; all having substantial challenges with day-to-day activities (Disability Prevalence Estimates 2011/12 (UK), 2012).

A systematic review examining definitions and operationalizations used in measuring prevalence rates of children with complex chronic health conditions found considerable variation in prevalence rates across countries. They attributed this inconsistency to the multiplicity in concepts and operationalizations used in terms of sources of information, the types of study populations and the retrieval of material used (Van Der Lee et al., 2007). They recommended the use of a comprehensive framework that utilizes international statistical classifications of diseases (ICD), in order to construct more consistent measurements. They also encouraged the development of international agreement on definitions of childhood chronic health conditions. However, one recent study found that in spite of the differences in diagnosis and case inclusion across databases, the construction of a common database for severe intellectual disability was deemed feasible through the synchronization of certain measures, such as age, and through restriction to those with severe intellectual disability only (Norman and Fraser, 2014). The lack of comprehensive data collection procedures poses a significant problem in the area of childhood disability research, thus making it challenging to estimate reliable prevalence rates for the development of services and planning for care in the future.

Much work is needed to improve protocols and procedures specifically in relation to children with LLC's in the community.

1.5.1 Prevalence of LLC's in Ireland and the UK

The lack of a uniform definition and poor data collection procedures have resulted in the research community reporting widely varying prevalence rates of children with chronic conditions (Newacheck and Taylor, 1992). In Ireland, approximately 1,400 children are living with a life-limiting condition, with up to 53% of deaths taking place within the first days of life, 15% in the first month of life and 32% within the first year of life (DOHC 2005). In Northern Ireland, a prevalence of 17.2 per 10,000 is projected (Laura Lynn, 2013). Ireland has experienced steady increases in the prevalence rates of children aged 6-8 years with severe intellectual disability; these were observed in a recent study comparing prevalence rates across Europe between 1990-2002 (Bakel et al., 2014) which is evidence of the rising life-expectancy of this population.

Up to 150,000 thousand families are caring for children with severe disabilities in the UK (Roberts and Lawton, 2001) and there is an estimated 12 per 10,000 children, aged between 0-19 years, with LLC's. Based on those figures, it is predicted that by 2021, there will be 1,610 children with LLC's in Ireland; however, this may be an underestimate, as it assumes Ireland has similar prevalence rates to the UK. Prevalence rates in the UK were found to be much higher than previously predicted; 32 per 10,000 (Fraser et al., 2012), as opposed to original projections of 16 per 10,000 (Association for Children with Life-Threatening or Terminal Conditions and their Families and the Royal College of Pediatrics and Child Health, 2009). One study conducted in Kent in the (UK) during the 1990's found a prevalence rate of 1.2 per 1,000 children living with a life-limiting condition (excluding malignancy) (Lenton et al., 2001). A more recent UK study, using time-trends and geographic locations to estimate prevalence rates of children with life-limiting conditions, found that the prevalence rate of children aged 1-5 increased by 27% (i.e. 8,400 – 10,700). In addition, they found a geographic gradient where the number of LLC's are highest in the more deprived areas and disadvantaged communities in the UK (Norman and Fraser, 2014).

Over the last three years, work has been done in Ireland to develop a national database to provide further information on prevalence rates. A combination of a 'Hospital In-Patient Enquiry' (HIPE) system and death certificate data was used to set up the database. Data is collected annually, by the health intelligence unit at the HSE and aims to inform service and workforce planning and the mapping of service delivery (DOHC 2010). To date, however, little progress has been made and data is still not available to the general public.

1.5.2 International Prevalence Rates

A projected 7 million children would benefit from paediatric palliative care services globally (Children's Hospice International, 2011). In Canada, more than half a million children younger than 20 years of age have some form of disability (Anderson et al., 2007) and as many as 3,889 children could benefit from involvement with a paediatric palliative care service annually (Widger et al., 2007). In the United States (US), 8,600 children would benefit from palliative care services (Field and Behrman, 2003). In Canada and the US combined, it is estimated that up to 31% of children have one or more chronic conditions (Newacheck and Taylor, 1992).

1.5.3 Mortality Rates

In Ireland, an average of 423 child deaths occurred annually between 2005-2010, and of these, 323 died from one of the LLC's outlined in the ACT categories shown in Table 1.1 (Laura Lynn, 2013). Deaths of children with life-limiting conditions account for 50% or greater of all child deaths in England and Wales; in particular, perinatal and congenital conditions are the most common cause of death (Sidebotham et al., 2014). Between the years of 2010-11, 968 children under the age of 18 years died as a result of chromosomal, genetic, and congenital heart abnormalities in England (Fraser et al., 2014). According to a study conducted in Wales, 54% of a total of 1,052 childhood deaths were caused by a life-limiting condition between 2002 and 2007 (Hain et al., 2013). The five leading causes of death were Cerebral Palsy (22 deaths), Malignant Neoplasm (15), Acute Leukemia

(11), Epilepsy and Muscular dystrophy (9). Another study assessing prevalence rates of children with LLC's in Wales proposed that prevalence is 10 times that of mortality (Hain, 2005). Annual mortality rates for children with complex chronic conditions declined by 33% between the years 1979–1997, according to one longitudinal study in the US (Feudtner et al., 2001). Of the 1.75 million deaths, which occurred among 0-24 year olds, 20% of non-cancer deaths occurred during childhood and 7% during adolescent years.

1.6 Palliative Care Policy in Ireland

Over the last fifteen years, a number of important policy documents and national reports were published, detailing recommendations for the development of both adult and children's palliative care services in Ireland (DOHC 2010, DOHC 2005, DOHC 2001, Laura Lynn, 2013). The most significant publication was *The Report of the National Advisory Committee on Palliative Care (NACPC)* (DOHC 2001). Adopted as a national policy in 2001, this seminal report made several recommendations for the planning, funding and development of specialist palliative care services, including the adoption of a comprehensive policy framework for palliative care. The NACPC outlined three distinct levels of palliative care and recommended that palliative care services should be organized around these levels of growing specialism, as presented in Figure 1.1 below.

Figure 1.1 Levels of Palliative Care

Level 1	Palliative Care Approach	Involves engagement in the principles of palliative care, where required, by all health professionals.
Level 2	General Palliative Care	Viewed as the intermediate level of expertise, with engagement in palliative care being part of the health professional's caring role but not defining it.

Level 3	Specialist Palliative Care	Whose practice involves palliative care as its core activity and is directed towards caring for parents with complex and demanding palliative care.
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Although the focus of this report was on adult services, recommendations for the development of a children’s palliative care service were made. It was agreed that home was the most appropriate place to provide palliative care, given that families are closely linked to a GP, public health nurse and a specialist palliative care team. A need for a review of palliative care services for children was acknowledged and the four ACT & RCP&CH definitions of children’s palliative care were adopted (as described in section 1.4.3).

In 2005, a subsequent needs assessment followed; A Palliative Care Needs Assessment for Children (DOHC 2005). This report was a joint collaboration between a team of researchers at University College Dublin (UCD), The Irish Hospice Foundation (IHF) and the Department of Health and Children. Despite significant progress since the previous palliative care strategy in 2001, numerous continuing issues in the provision of care services for children with LLC’s were highlighted. Several recommendations were made including the need for more partnership between statutory and voluntary organizations, which would provide more security and responsibility in their roles. An urgent need for more specialized training in paediatric palliative care and the creation of more specialized medical and nursing posts were outlined. Training for families was also viewed as essential to enable them to care for their child at home. It was also agreed that the development of home-based services, i.e. respite care, was a priority area. The development of individualized care plans to be implemented by a “key-worker”, who acts as a liaison between families and all services, was also seen as essential.

In 2010, the first national policy, Palliative care for children with life-limiting conditions in Ireland - A national policy, was published (DOHC 2010). Echoing the same issues highlighted in the needs assessment, several new policy objectives were outlined under the principles of inclusiveness, partnership, comprehensiveness and flexibility. An implementation plan for the appointment of a

paediatric palliative care consultant, a number of regional outreach nurses and children's palliative care team was delineated. The establishment of a committee for children's palliative care, to provide a national forum for interconnected development on children's palliative care services, was proposed. In addition, the development of a national database was also given priority and the importance of clinical governance in the management of care in the community was advocated.

In 2013, a report, *Respite Services for children with life-limiting conditions and their families in Ireland - A national needs assessment*, was published (Laura Lynn, 2013); a joint collaboration between the Irish Hospice Foundation and Ireland's out of home respite care provider, Laura Lynn. This report provided an assessment of the progress made on the policy objectives set out in 2009's National strategy. The HSE's limited coordination and direct delivery role in the provision of respite care for families were highlighted. Key recommendations included: a need for national standards to be developed by the National Advisory Committee for Palliative care (NACPC) and an assessment tool for respite services to be examined. Regional groups representing paediatrics, disability services, acute and community services, children's outreach nurses and specialist palliative care services should be used to inform the provision of services and ensure the implementation of national standards. In addition, it was recommended that the health service work closely with both voluntary and statutory service providers on whether facilities already in place need further development. Finally, the educational needs of organizations nationally and regionally were stressed as important considerations in developing and furthering health care professionals' knowledge in palliative care. An estimated budget of 10-12 million would need to be invested by 2021 to ensure these objectives are acted upon.

In terms of progress from these policy directives, recent analysis of Ireland's National Palliative care strategy for adults found that policy initiatives in a relatively new area of healthcare could face trade-offs between ambition and feasibility (May et al., 2013). The policy objectives outlined for children with LLC's (DOHC 2010) clearly state that the objectives must be met using existing resources. This is challenging, given that over 2 billion euros has been taken from public expenditure between 2010 – 2012; 17% of which was taken from the health care budget (Thomas et al., 2012), so progress is inevitably hindered. However, in the area of children's palliative care, some progress has been made with the

appointment of a consultant paediatrician, with a special interest in paediatric palliative care, and eight clinical outreach nurse specialists. Education and training courses have improved and more specialized training is taking place, however, much work is needed to develop services nationally. This extensive study will provide important insights into the service experiences to date and families' preferences of care.

1.7 Profile of Children in this Study

The children in this study are aged between 1 and 7 years of age. The age of the children in this study is largely determined by the organisation with which we chose to conduct this study. The organisation traditionally provides care to children up until four years of age, but in some cases has continued to provide care up to seven years. The children in this study have complex life-limiting neurodevelopmental conditions and have significant care needs. All children in this study were cared for at home by a parent (predominantly mothers) on a full time basis. The range and complexity of the children's needs are discussed in chapter 4.

1.8 Current Service Provision

In Ireland, care for children with life-limiting conditions is primarily provided in the home, with parents typically fulfilling the role of primary carers. Several statutory and voluntary organizations are involved in the provision of care; The Jack & Jill Children's Foundation, Laura Lynn House, Suzanne House, St Michaels and St. Catherine's are all examples, with each one varying in their capacity to provide care. In addition to those, diagnosis specific organizations, like Down syndrome Ireland and other parental support groups provide various forms of support, and in some cases, provide funding towards the additional costs of care. Examples of the additional supports provided by organizations include: in-home respite care, out of home respite care, home help, physiotherapy, speech and language therapy and occupational therapy. In 2011, Ireland's first children's palliative care hospice was established 'Laura Lynn House' and is a dedicated 8 bed children's palliative care unit. In the case of in-home respite care, where it is possible, care is provided by a

registered general nurse or trained carer. The numbers of children at home in Ireland is unknown and not easily identified due to the overlap of children with disabilities and life-limiting conditions (Laura Lynn, 2013). Existing agencies do provide some insight; the Jack & Jill Children's Foundation provided care to up to 280 families in 2012. It is not known how many families in total receive statutory services.

1.9 Overview of Study

The following section presents the rationale for conducting this study and Figure 1.1 presents an overview of the study aims and objectives of phase one and phase two and the final outputs of the study. The chapter concludes with an outline of the format of the remainder of the thesis. A more detailed discussion of the study objectives is presented in Chapter 2.

1.10 Rationale for Study

Children with complex care needs typically, (i) require substantial family-identified health care service needs, (ii) have one or more chronic conditions, (iii) have severe functional limitations, which can often require assistance from technology, and (iv) have a high utilisation of health care services (Cohen et al., 2011). Using services is a significant part of the daily lives of parents caring for children with LLC's, as found in one study where hospital doctor appointments ranged from 1 to 104 visits in the previous year (Sloper and Turner, 1992). Navigating a myriad of care services is challenging for parents (Hunt et al., 2013b) and their views on such experiences can provide valuable insight into the development and planning of future care services.

In Ireland and the UK, many services are based on the views of health care providers and not the experience of those service users involved. Scant evidence exists on the incorporation of child and parent views into the decision making and future planning of care services (Noyes et al., 2013), yet it is an important and useful practice, as demonstrated recently in the UK's first in-depth study on care needs of over 1,000 children with LLC's (Hunt et al., 2013b). Developing care services to match children's care needs is currently a national priority (DOHC 2005,

DOHC 2010, Laura Lynn, 2013, Grinyer, 2011) and the needs of children with LLC's is recently receiving more care and attention; however, policy recommendations are broad and attention to unique needs are minimal or absent (Rushton and Catlin, 2002).

1.10.1 Outcomes of the Study

This study provides a comprehensive evidence base that can be used to inform Ireland's health policy objectives for children with life-limiting neurodevelopmental conditions. It incorporated the views of service users by 1) examining parents' experiences of using services on behalf of their child, 2) using a stated choice method to identify preferences for different components of care packages and 3) defining the financial impact on families caring for children with LLC's under the current government policy. This small population experiences an important and increasing set of problems and their needs are complex and multifaceted. This study provides new evidence that clearly defines parents' experiences, preferences of care and the financial implications of caring. To the best of our knowledge, this is the first study to apply discrete choice experiment methodology to this population and as a result, it provides a unique and important contribution to the literature. Equally, little evidence exists on the out of pocket costs associated with providing care to children with LLC's at home and the findings from this study highlights the significant financial impact experienced by families. It is hoped that this research will contribute to the significant research agenda outlined by (Quinn and Bailey, 2011, Emond and Eaton, 2004, Ling, 2012), as the future planning of services for children with LLC's is a critically under-researched area (Noyes et al., 2013).

Figure 1.2 Overview of study Objectives

Aim: Provide a detailed and reliable evidence base on the health care experiences and service preferences that relates to children with neurodevelopmental life-limiting conditions and their families at home.

Objectives:

Examines parents' experiences of caring for children with life-limiting disabilities.

Elicit parent's preferences and willingness to trade for an optimal monthly care

Examine parents out of pocket costs associated with caring for their child

Methods:

Interviews
(Phase 1)

Quantitative questionnaire
/ survey (Phase 2)

Outputs:

- (1) Identification & explication of parents' care experiences and service needs.
- (2) 8 important dimensions of need identified

- (1) Identification of specific service preferences & willingness to trade between various services.
- (2) Identification of the financial costs of caring

Findings:

Chapter 4

Chapter 5 & 6

Chapter 7: Discussion of key findings from all stages of the study

Chapter 8: Conclusions and Recommendations

1.11 Format of the Study

Chapter Two presents a critical appraisal of the research literature, which relates to children with life-limiting disabilities. Gaps in current knowledge are highlighted.

Chapter Three provides a detailed overview of the research design, recruitment criteria, profile of participants, sampling frame, and ethics.

Chapter Four presents the qualitative study, namely, Interpretative Phenomenological Analysis; the design, methodological orientation, data collection and analysis are presented, along with the findings and a preliminary discussion. The strengths and limitations of the study are also outlined.

Chapter Five presents the quantitative study, namely, a Discrete Choice Experiment; the design, methodological orientation, data collection and analysis are presented along with the findings and a preliminary discussion. The strengths and limitations of the study are also outlined.

Chapter Six presents a descriptive study on the out of pocket costs of caring for a child with a life-limiting condition and some free-text commentary provided by parents.

Chapter Seven discusses the findings from the three studies in detail and future directions are proposed.

Chapter Eight discusses the overall strengths and limitations of the study; the implications for relevant stakeholders and recommendations are made for service planners and policy makers.

Chapter 2

The Literature review

2.1 Introduction

This chapter presents a critical review of the research literature related to this study. This includes a review of the literature that relates to caring for children with life-limiting conditions. Relevant gaps in the literature, which relate to the aims of the study, are highlighted.

2.2 Terminology

We provided a brief discussion on the issues around definitions and the terminology used to describe children with life-limiting conditions in chapter one. The terms used vary between “life-limiting, palliative care”, “life-threatening” and “complex needs” and these encapsulate a broad spectrum of diagnoses. Other challenges include the lack of a clearly defined population in the research literature; some studies have included a diverse population, with a wide range of illnesses, and others are strictly defined to one particular illness category. This posed challenges when deciding what research studies to include in the literature review. Outlined below is the inclusion criteria and underlying rationale it.

2.3 Inclusion criteria

All of the children in this present study have life-limiting conditions and, in most cases, a specific neurodevelopmental diagnosis. As already discussed above, the issues around terminology present some challenges in identifying and defining this patient population within the research literature. One defining feature, however, is the fact that children with LLC’s have complex needs. The common characteristics regardless of diagnosis are outlined below and were used as a guide for what to include in the literature review:

- Children who have a diagnosis of a life-limiting condition

- Have on-going medical needs
- Have dependence on some form of medical equipment / technology

Particular emphasis was given to the non-malignant paediatric populations but this was not strictly applied, given that some studies included a combination of both malignant and non-malignant populations. Finally, where possible, studies were sourced that included populations similar in age to the children in this study for generalisability but this was not strictly adhered to given that some of the larger scale studies included children up until the age of 18 years. All of the criteria outlined above was applied to the literature review which was broad and expansive.

2.4 Search strategy

A critical review of published literature was undertaken during the course of the study between 2011-2014, with periodic reassessment conducted throughout. In reviewing the literature, electronic databases, including, Cochrane Library, Medline, Proquest, Blackwell, PsychArticles, ScienceDirect and Synergie were used. Searches were undertaken on major texts and policy documents from the Irish and UK government. Literature from voluntary organizations were reviewed, including the Association for Children with Life-Limiting or Terminal Conditions and their Families (ACT), Irish Association for Palliative Care (IAPC), Irish Hospice Foundation (IHF), as these were official government publications relevant to palliative care, children with life-limiting conditions, and children with disabilities. Websites and search engines used included Google, and Google scholar. Key search terms included “life-limiting”, “life-threatening”, “complex needs”, “disability” and “neurodevelopmental disability” and “home care” “respite care”, “models of care”, “services”, “community care”, “hospice care”, “palliative care”, “impact on carers”, “financial challenges”, “mothers experiences”, “parental perspectives”, “coping”, “care needs” and “preferences”. Searchers made use of broad terms, Boolean (and, or) and truncated terms. No date restrictions were applied to the literature search and all literature included covered the years, 1980-2014. The search was confined to English language material only.

2.5 The literature – An Overview

This chapter presents a critical review of the literature related to children with life-limiting conditions and is presented in three main strands. The first of which includes a review of the nature and experiences of caring for a child with a life-limiting condition. The second section includes a review of the impact on families and the coping strategies and resources used to ameliorate their situation, while the third section includes a review of the financial costs of caring. The literature is critiqued at each stage and its relevance to the population in this study is discussed. Relevant gaps and limitations are highlighted, which are related to the aims of the present study.

2.6 The Nature and Experiences of Caring

Kirk and Glendinning (2002) examined the experiences of providing home-care to children with technology dependency in the UK and found that parents had no choice but to become expert carers, which required administering medical procedures and making expert judgment on their child's requirements, where the family's lives revolved around the technology in the home. They also reported that parents invested a significant amount of time advocating for services on behalf of their children. Support through a regular baby sitter could not be obtained due to the complexity of the child's illness, which compounded an already stressful situation for the primary carer. Work by Redmond and Richardson (2003) explored the service needs of Irish mothers (N=19) with children who have severe to profound disability. They described parents as quasi-nurses, which relates to the type of care required by their child, namely, the administration of complex medicine and the maintenance of technically sophisticated equipment and procedures. The findings from this study also revealed that children with complex life-limiting non-malignant conditions fall between two major service providers, the acute hospital system and services for those with intellectual disability. The families who do not easily fit into a recognisable care category may be excluded from receiving essential services.

Another Irish study by Nicholl and Begley (2012) investigated mothers' (n=17) caregiving experiences of caring for children with complex needs in Ireland. Four caregiving categories emerged from the analysis: 1.) *Normal mothering*, which includes diaper changing, dressing, toileting etc., is sustained when normal milestones are not achieved, resulting in care becoming more demanding; 2.) *Technical caregiving*, which includes medication management, mobilization, seizure control and the management of medical technology including suction machines and feeding pumps, with parents often emulating skills of health care professionals. In addition, mothers also carried out physiotherapy and speech and language therapy to progress their child's health; 3.) *Preemptive caregiving*, which was a skill developed by mothers as a result of their expert knowledge of their child's condition. Mothers put measures in place to prevent their child's deterioration, which required them to constantly think ahead and forecast potential problems; 4.) *Individualized caregiving*, which relates to managing the individual behaviour of the child, which, in the case of children with complex illnesses, can be unique and challenging. Mothers are intensively tuned into their child to observe signs of distress, so they can provide comfort and safe guard them from challenging situations.

A phenomenological investigation by Ware and Raval (2007) examined fathers' (N=8) retrospective experiences of caring for children with life-limiting conditions in the UK. Findings revealed that fathers struggled to acclimatize to their "changed world" and felt the pressure to be strong for their partner and family. They found it difficult to express their emotions in comparison to their partner but also in the presence of other fathers in similar situations. They expressed the importance of looking after themselves because they felt that being depressed would have an adverse impact on the whole family; one father suggested taking up boxing or a pastime that would help to alleviate stress. Fathers found contact with families in similar circumstances useful for information sharing and discussions on practical matters. Engaging with support groups and concentrating on what they could do to help their family; financially supporting them and actively engaging in campaigning and petitioning for supplementary services were all helpful coping mechanisms.

A grounded theory study by Steele and Davies (2006) on the impact on parents (N=8) and families caring for a children with progressive, life-limiting conditions described the emotional fear, uncertainty and grief that formed the fabric

of their everyday lives. Emotions were always present and fear and uncertainty were exacerbated when their child's condition worsened. Emotional stress faded as parents became more familiar with their situation but the physical impact expended energy, as among the caring activities engaged in by parents included advocacy, preparing restricted diets, attending medical and therapy appointments, all while trying to maintain their child's health. Spiritually, parents expressed experiencing deep spiritual pain and stated that they searched for answers as to why this had happened to their family; most parents acknowledged a religious affiliation and in some cases asserted that it had helped them to cope with the situation. Roberts and Lawton (2001) extensively reviewed 40,000 records on families caring for disabled children and found that severely disabled infants (<12 months) require additional clothing and washing due to recurrent vomiting or diarrhoea. Also, the complexity and large time investments were required as a result of infants being excessively floppy, rigid and attached to machines. Extra care for the infants included; feeding the infant through a tube, administering medication and interrupted sleep that comes when the infant (in one case reported) was constantly crying through the night. The intensive nature of caring was further amplified by the fact that these infants cannot be left alone because of the danger of fits or choking.

A Canadian study by Carnevale et al. (2006), using an interpretative framework, examined parents' experiences of caring for ventilated assisted children at home. Findings revealed that parental responsibility was found to be stressful and overwhelming because of the considerable time investments of constant care and attention required by their child. They struggled and experienced emotional strain because of the physical and psychological dependence of the child. Seeking normality required large efforts and routines were strictly adhered to, so they could mimic a 'normal family'. Families were offended by the social reactions they received and felt isolated, particularly because neither their family nor the medical system could support the families respite needs due to the complexity of their child's condition. In Ireland, care is provided in the home and services are community based (DOHC 2010, DOHC 2005).

In summary, the review of the nature and experiences of caring reveals that caring for a child with a LLC at home is multifaceted and requires significant input from families, health care providers and health care systems, but in most cases, the emotional and physical burden is absorbed by families. Huge time investments are

devoted to carrying out complex tasks and the constant monitoring that is required. Isolation is commonly reported and fathers in particular report feeling the need to remain strong for the family. Some limitations regarding the literature review warrant mention, in particular; the heterogeneous populations used in the studies make the results less generalizable to children with life-limiting neurodevelopmental conditions. The small sample sizes in some cases, although typical in most qualitative studies limits the recommendations that can be made. Also, more research is needed to examine fathers' experiences and perspectives, as this has the potential to highlight important differences of the ways men cope with their child's condition in comparison to women. One of the goals of this study was to include fathers as much as possible in the research process by encouraging them to participate in the interviews. Overall, the literature reviewed shows consistencies across diagnostic categorisations, where, universally, parents are dealing with an ongoing chronic situation involving a plethora of stressors arising from different situations and predicaments, which suggests similarities exist in the challenges experienced across a broad spectrum of diagnoses.

2.7 The Medicalised Home

Complex nursing care, involving highly technical procedures, are now being carried out in the home which places enormous physical and emotional demands on parents (Watson et al., 2002). Technological advances and improvement in survival rates increases the need for care to be provided in the home. Home-care is defined as “a return to a system and set of priorities in which family values are important to the care of a child with a chronic health problem as they are in the case of the well child” (Stein, 1985 p.90). Work by Strickland et al. (2004) describes home care as a complex balancing act and highlights important goals in relation to successful home care, which include, normalising the life of the child, minimizing the disruptive impact of the child's condition on the family and fostering the child's maximal growth and development. They also define the necessary elements of successful home care: 1) the family want to have their child at home, 2) the family is able to cope with the intrusions and stressors imposed and 3) the family receives quality supportive services. Strickland et al. (2004) suggest that coordinated care may be

the element that most distinguishes the medical home from traditional paediatric practice and that this important element is lacking due to families failing to connect with discharge planning teams, who assist in the successful transition from hospital to home. Similarly, Arras and Dubler (1994) highlight that some families are discharged home without any training or adequate instruction and are often ill-prepared for the magnitude of their role. Oldman and Beresford (2000) suggest that a new way of thinking about domestic environments and disability may mean that the definition of housing needs go beyond stair lifts and ramps to issues of poverty and health and the constraints of physical space. Space constraints present additional challenges for families, as the presence of heavy equipment, special chairs and walking equipment all serve as potential hazards and provide less space for children to move around and progress.

The American Academy of Pediatrics (2002) points to the fact that efforts to establish medical homes for all children are hindered by the lack of reimbursements, geographic locations, personnel constraints and social forces. They outlined the successful components of comprehensive and effective health care at home: accessible, family centred, continuous, comprehensive, coordinated, compassionate and culturally effective. The numbers of children receiving care at home in Ireland is unknown and difficult to estimate due to poor data collection mechanisms, however, Roberts and Lawton (2001) estimate that 150,000 UK families are caring for severely disabled children which provides a possible approximation for Ireland. The review of the medical home provides a clear description of how families' lives change, especially in relation to the intensity of caring and the disruption the technology imposes on family living environments. Space is reduced and adaptations are necessary to accommodate their child's needs.

2.8 Impact of Caring

2.8.1 Impact on Carers

Parents caring for technology dependent children at home experience a wide range of emotions such as anxiety, anger, guilt, frustration and sorrow (Patterson et al., 1992). Breslau et al. (1982) examined the psychological distress in mothers

(N=369) and found that the greater the disabled child's dependence in daily activities, the more significant the psychological distress that mothers experienced. Additionally Thyen et al. (2003) found that parents are at risk of depression to the extent that requires medical attention. Feelings of isolation and depression are commonly reported throughout the research literature and intervention programmes aimed at promoting and supporting parents mental health are needed (Emond and Eaton, 2004). A Scottish community survey by Hoare et al. (1998) examined psychological adjustment, carer distress and the effect of respite on families caring for children with severe intellectual disabilities and found that mothers with qualifications reported much higher scorers on several indices of stress, which agrees with findings from Wishart et al. (1981), where mothers with higher levels of education reported greater distress. Both studies concluded that this group of mothers may have a more realistic assessment of their child's situation.

2.8.1.1 Impact on Family

Relationships between family members can become strained when caring for a child with a life-limiting condition (Hunt et al., 2013b). Many studies have researched the specific effects, including a widely cited UK study by Quine and Pahl (1985), that investigated the causes of stress in families caring for severely disabled children. Findings revealed the most predominant causes of stress includes behaviour difficulties in relation to their child, night-time disruptions, social seclusion, hardship in the family, the wide range of impairments and financial concerns. Families' experiences of living with long-term technology dependent children have described a life where the balance is constantly changing and unpredictability prevails, with parents having limited control due to the uncertain nature of their child's illness (O'Brien, 2001). Parents report having difficulty maintaining quality in their relationships and lamented on the lack of time they have for each other now that their child is their number one priority. In addition, some families made a firm decision to limit family size as a result of having a technology dependent child, whereas other families made a conscious decision to have additional children (O'Brien, 2001).

A recent qualitative study in the UK by Whiting (2014) examined differences in the need for help and support in three sub-groups of (N=33) families that had either, 1.) a child with a disability, 2.) a child with life-limiting or life-threatening condition or 3.) a child that is technology dependent. Three core themes relating to family life were consistent across the three subgroups:

- 1.) *Time*: This is where families considered themselves ‘time poor’, as many of the following tasks required considerable time investments; helping their child with daily activities of living, preparation and administration of medicines, moving and stretching exercises, providing technical care such as using suction machines and oxygen delivery systems, attending hospital appointments and keeping the house tidy for medical professionals to visit their home. All of these tasks reduced their available time for other family members.
- 2.) *Multiple roles*: This was the second core theme, where parents described being a parent, carer, physiotherapist, nurse and taxi driver all in one, and although this role does not differ significantly from a typical maternal role, parents had to conduct these roles alongside the additional tasks listed above. Parents expressed concerns over the dichotomy of being both a parent and a technical care provider
- 3.) *Disabled Family*: The third and final theme is the disabled family where the impact of their child’s diagnosis impacted upon other parts of family life such as social disengagement, loss of employment opportunities and a general disruption of the household and family life; families describe their lives as dysfunctional as a result of their child’s illness.

2.8.1.2 Impact on Siblings

The psychosocial effects on siblings of children with autism was examined by Bågenholm and Gillberg (1991) and findings revealed that siblings were more likely to feel lonely and have concerns about the future. In addition, they regarded their siblings to be a burden. The extensive literature review on siblings perspectives by Knecht et al. (2014) revealed the considerable emotional impact on siblings, where they are often withdrawn and stay in the background; as there is an extensive focus on the sick child. They receive less attention from parents, family and friends and as

a result they feel lonely. Conflicting emotions were also found, where they exhibited feelings of jealousy and envy and felt that an injustice was imposed on their family. Other findings reveal eating changes, weight changes and sleep disturbances, however, many of the studies reviewed by Knecht et al. (2014) were from a proxy and not from the siblings themselves. They called for further research on siblings' perspectives to make their voices heard. Additionally, Emond and Eaton (2004) calls for the examination of optimum coping strategies that may support and influence the well-being of siblings.

2.9 Key Areas of Unmet Need

A national survey by Beresford (1995) on the care needs of children and their families in the UK (N=1,061) found the most common unmet needs were financial support to pay for additional costs incurred. Parents expressed a need for additional financial support and a preference for a weekly cash payment in contrast to an annual grant payment, as frequent payments would be more beneficial. Other needs included aid in the planning of their child's future, help to support caring for their child, and information on available services. Another UK study by Sloper and Turner (1992) investigated the service needs of families (n=107) of young children with severe disabilities at two key time points, the initial stage of diagnosis and when the child commenced school. Their statistical analysis at both time points revealed that families had engaged with an average of 10 professionals in the previous 12 months; the most frequently visited were physiotherapists, teachers and hospital doctors. Hospital doctor appointments ranged from 1 to 104 visits in the previous year, indicating not only the intensity of service use but also the multiplicity of different agencies involved in the care of their child. Only half of the respondents reported having a link person to help with accessing services at both time points and had to acquire information and coordinate services by themselves. One third of families had greater than 13 appointments with physiotherapists in the previous year, indicating on average one session per month, which is similar to Ireland. Teachers and Physiotherapists were rated as very helpful by parents and were the most valued out of all service types listed. Some of the most important perceived needs expressed by parents were the need for regular consultations on

their child's development, information on services, improving their child's mobility and information regarding their child's condition. The largest area of unmet need reported was the provision of information about services for their child. More than half of the parents in the study reported not receiving any information and how they would like additional communication on services, which is similar to findings by Emond and Eaton (2004) where information provision is a key issue for parents and carers.

A UK study by Horrocks et al. (2002) evaluated a service aimed at providing nursing and psychology support in the community to families (N=29) caring for children with non-malignant life-threatening conditions, echoes similar findings to those outlined above where parents would like an improvement in communication on services available to their child. Respite care represents a crucial support for parents, as it allows them to take some well-needed time off to rest or conduct other important activities in the family home. One study examining respite care for children with life-limiting conditions found that parents are hesitant about giving their child over to health care professionals that do not have experience caring for children with life-limiting conditions. Ling (2012) also found that trust was an important element in improving parents' confidence in using respite care. Noyes et al. (2013) believes future planning of care services for children with LLC's is a critically underresearched area. Their research on parents and children's evaluation of an information tool "my choices booklets", revealed deficiencies in the following areas; limitations in the availability of information, lack of shared knowledge and planned elicitation of child and family care preferences. Families called for better signposting of information and the study concluded with recommendations for staff and nurses to receive further training and support to actively engage with care planning tools such as the "my choices booklet".

Children with complex conditions typically require technical and or medical equipment in the home. Examples of technology include; gastronomy, nasogastric feeding device, tracheostomy, oxygen, suction and ventilator support (Beale, 2002). A recent Irish study shows that these children can require up to 22 pieces of equipment to support their conditions (Nicholl et al., 2013). Children vary in characteristics of technology dependency. The difficulty of provision and funding arrangements for technological/medical equipment continues to be challenging for families, where they are required through the necessity and lack of support to adopt

the role of co-ordinating equipment and supplies in order to provide suitable and safe care for their child (Watson et al., 2002). Reeves et al. (2006) reports that technology strips parents of their role as a parent and that the combined roles of performing nursing care and being a parent is challenging, as parents need to feel like parents and not nursing aids. Moreover, Winkler et al. (2006) point out that the meaning of 'home' is altered by the intrusion of medical equipment in the home. Issues around the unreliability and poor supply of equipment was reported by Kirk (1999) where the ordering of equipment could be forgotten and the wrong equipment ordered. Parents collected large supplies of heavy equipment and stored them at their homes which was sometimes a source of tension with district nurses and health visitors, as they felt parents failed to understand that they could not predict the length of time their child would need equipment. Funding disputes arose and was described as a nightmare by community nurses, who spent significant amounts of time sorting these problems.

Not all of the children in this present study had technology dependence. Wang et al. (2004) differentiates between the technology dependent child and the child that has chronic disability and adopts the definition of technology dependence as, 'a medical device to compensate for the loss of vital bodily function and substantial and ongoing nursing care to avert death or future disability' (Office of Technology Assessment, 1987 P.3). Research on the impact of medical technology on children and families at home is lacking and has to date been inadequately investigated. Evidence is needed for the successful implementation of children and families to return to the community and sustain quality of life (Wang and Barnard, 2004). The key areas of need in relation to caring for a child with a life-limiting condition include; information, respite care and support in procuring equipment. Physiotherapists were rated highly by parents, as was a weekly cash payment towards the additional costs of caring. Trust is an important element in using services and support for families in planning ahead for future care needs was seen as essential. The experiences have been investigated from various view-points, but more large scale studies are needed; also, many of the service needs may not be generalizable due to the different health care systems, however, the studies based on the Irish context share similarities with the other studies reviewed.

2.10 Stress and Coping Strategies

The most widely cited model of stress and coping is that of Lazarus and Folkman (1984) where individuals are viewed as actively and creatively seeking to reduce stressors as they are encountered. Coping is defined as “the process of managing demands (internal or external) that are appraised as taking or exceeding the demands of the person” (Lazarus and Folkman, 1984 p.283). More recently a revised model of stress and coping has emerged which places more emphasis on the positive psychological states in the coping process (Folkman, 1997). An extensive review by Beresford (1994) on the resources and strategies used by parents to cope when caring for a disabled child indicates that no one coping strategy can be considered good or bad, adaptive or maladaptive and that accounts need to be taken of the nature of the stressor, the availability of coping resources and the outcome of the coping effort. The study also found that “controllability” is an important feature of the nature of the stressor and that a problem-focused strategy would be most useful to families to manage controllable stressors. An example provided is where parents use over-night respite care to help with over-tiredness. If the stressor is uncontrollable, she suggests using an emotion-focused strategy where the parent focuses for example on the educational rather than physical achievements of their child. Beresford (1994) extensively reviewed research on coping strategies used by families including the dynamic work of Bregman (1980) where the researcher lived for 96 hours with six different families whose children had progressive neuromuscular diseases. The aim was to identify parents’ positive coping strategies when caring for their child. Positive examples uncovered include a “take each day as it comes” philosophy where parents focused on the present. Parents encouraged their children to spend time in the company of healthy children, remained well informed and actively engaged in educating community members about their child’s condition. Finally, parents engaged in recreation to release emotional strains and released emotions through crying.

Other research by Brown and Hepple (1989) show talking to one’s spouse about their situation, accepting help from extended family supports, and focusing on the positive aspects of their child were all found to be useful coping strategies. Research shows that high use of the coping strategy, ‘passive optimism’, where mothers take an inactive approach to obtaining help and support, reduces their

likelihood of obtaining services to more suitably meet their needs (Sloper and Turner, 1992). Advocacy and reframing are useful strategies employed in increasing stability in family life as found by O'Brien (2001) where families continuously engaged in activities to advocate on behalf of their technology dependent child. In addition, reframing involved focusing energy on achievable outcomes and re-examining beliefs and values and prioritizing what they felt was important. The literature on coping strategies although dated provides insight into the useful activities parents can engage in to alleviate the stress of caring for their sick child. A focus of this present study is to identify the forms of emotional support services needed by parents caring for children with LLC's. Although only a small focus of this present study, it will serve to inform service providers on what parents' need and how much they value support services.

2.10.1 Positive Experiences

Parents encounter many stressful situations when caring for a child with a LLC and the way parents cope with such situations can be an important indicator of well-being. There is an emerging literature on the positive aspects of coping, for instance, making positive comparisons to their child's past health and that of the present was found to be helpful for parents, and in particular, observing family growth and the adaptations they made over time helps parents to find sense and meaning in their experiences of caring for a technology dependent child (O'Brien, 2001). Another emerging concept in coping with adversity is posttraumatic growth, which is defined as growth occurring "as a process of struggling with adversity" (Tedeschi and Kilmer, 2005,p.124). A recent study by Cadell et al. (2014) on posttraumatic growth found that parents who have the highest scores of wellbeing, as reflected by higher self-esteem, optimism, spirituality and lower depression, may find it easier to find meaning in their situation Other research examined the cultural differences on the positive impact of having a child with a disability between Anglo and Latino mothers at different time points (Blacher et al., 2013); in this study, they found that Latino mothers had higher scores on positive impact on all measures in comparison to Anglo mothers. Although previous research has focused on the negative aspects of caring, more recently, there has been a focus on identifying the positive aspects

of caring for a child with a disability, where parents experience a change in priorities and perspectives, as a result of having a child that is unwell.

2.11 A Review of Service Models

Sloper (1999) extensively reviewed a number of service models that are effective in covering a broad range of parents' needs, the first of which is the "key worker" model (also known as link-worker), where a named person can be approached by parents for information and advice about a problem related to their child (Sloper, 1999). Their role requires continuous contact with parents and professionals to liaise and co-ordinate services for families. The key worker model has received notable attention in recent years in Ireland and is now widely recommended (DOHC 2010, Laura Lynn, 2013, DOHC 2005), with investment planned for key workers to be assigned to citizens with complex care needs in the community (Health Service Executive, 2014). Sloper and Turner's (1992) UK study examined the service needs of families (n=107) caring for a severely disabled child and described the role of the key worker from the parents' perspectives. Parents valued being able to contact a key worker with several problems related to their child, particularly when seeking information on services, emotional support, consistency of contact and helpful advice on how best to support their child. Multivariate analysis indicated that a key worker model was more valuable to parents with the greatest difficulties and less internal family resources (Sloper and Turner, 1992).

Again Ware and Raval (2007) research on fathers' experiences of caring for a child with a life-limiting condition found that the number and intensity of health care appointments related to their child overwhelmed fathers. Fathers suggested that one person (a key worker) would be an excellent type of support for parents to contact on all aspects relating to their child's care. Work by Appleton et al. (1997) highlights the importance of mandating front line clinical and administrative staff, so a full commitment is involved in the key worker role. They stressed the importance of the key worker being able to communicate easily across a supportive, intra-agency, client focused environment.

Sloper (1999) reviewed three additional models of care:

- 1) *Parent counselling model*: although more common in the USA, has the role of exploring a broad range of matters raised by parents, including issues around resources, relationship difficulties and challenges with their child. They help families to set goals and develop plans to carry them out. Feeling supported by professionals decreases stress around the child and can improve the child's development.
- 2) *Parent partnership model*: this focuses on partnership between parents and professionals where the emphasis is on both sides reaching a consensus on the needs of the child through open discussions and strong communication. There is a small but emerging body of research on the lack of congruence between parents and health care professionals on care needs (Blackard and Barsh, 1982, Bailey, 1987, Sloper and Turner, 1991, Garshelis and McConnell, 1993, Allen, 2014), so the parent partnership model may be a promising solution, given that parents call for their competence to be recognized by health care professionals (Neill, 2000, Hunt et al., 2013b).
- 3) *The coping skills model*: more common in American literature than the UK is where stress and coping theories are used to inform parent training in a myriad of skills from problem solving to accessing and utilizing social networks and coping strategies, like positive self-talk and relaxation. All of the 3 models were evaluated and after a two year follow up, mothers reported greater communication skills, enhanced coping skills and lower rates of depression (Sloper, 1999).

Parents consistently call for better quality information and communication in relation to the management of care for their child (Selman et al., 2009, Kirk and Glendinning, 2002, Almasri et al., 2012, Hunt et al., 2013b) and a need for one centralised information system. Policy frameworks in Ireland have largely emulated the UK, as they are far ahead in the development of care services for children with LLC's. A study by While and Dyson (2000) identified two model types in the UK: 1.) The community model, where there is strong connections to primary health care

and other local service providers and 2.) A hospital outreach model, with strong connections to the hospital service.

The models reviewed above provide an overview of the models of care that relate to children with life-limiting conditions. The key-worker model is most relevant to Ireland's care context and is the most established and widely recommended. Progress with the key worker model in Ireland is patchy, as implementation largely lies in small pockets of health care services for children with LLC's. Disconnect between the community and larger-scale multidisciplinary teams is a core challenge, thus, support is needed at all levels to exemplify this model and support its existence. Evaluating services for children with a life-limiting condition is challenging, as outcomes for this particular patient group are sadly more often mortality. According to Cass and Kugler (1999) developing a framework should involve the collation of information on specific needs from parents, referrals, purchasers and other involved professionals to determine the areas of conflict involved. There is little evidence available on the strengths and weaknesses of the different models reviewed in this section and much more evidence is needed to inform and develop suitable models for this patient population.

2.12 The Financial Costs of Caring

Research on the financial costs of caring for a child with complex disabilities has emerged over the years but there still remains significant gaps in the literature. A UK study by Dobson and Middleton (1998b) found that it was sixteen times more expensive to raise a child with a disability from birth to 17 years of age (£125,000) in comparison to raising a non-disabled child (£7,355). In the United States, Meyers et al. (1998) examined out of pocket costs for welfare recipient families caring for a child with mild to moderate and severe disabilities. Using household survey data in California, they found that unemployment rates were higher for mothers caring for more than one child or a child with a severe disability. Out of pocket expenses were higher for families caring for a severely disabled child; 15% (N=1,696) of families with severe disabilities had expenses greater than \$100.00 in the previous year, in comparison to 10.7% of families with mild or moderate disabilities. A concerning finding, was that families in receipt of welfare payments were still at a high risk of

experiencing hardship such as hunger, homelessness or eviction. Hunger was reported amongst 31–38% of families, including those with all levels of disability but it was more likely to be reported within families caring for more than one disabled child. O'Brien (2001) found, that despite families being covered by Medicaid, and in some cases private health insurance in the US, they still report having considerable expenses in relation to their technology dependent child. Examples include, paying for items not covered by private health insurance, such as having equipment constantly charging, as well as heating bills. One single parent in the study estimated that it cost 20,000 dollars in the previous year according to her taxes, and described exhausting all resources including borrowing against their pension. Research on the effectiveness of financial benefits provided to families is scarce and little evidence exists on the true impact of such supports. Beresford's (1995) UK national survey found that over one third of parents caring for a disabled child (n=1,061), with a higher proportion (9/10) of lone parents were surviving on benefits only, having no other sources of income. Estimates of welfare benefits paid to families were compared to parents' perceived minimum essential costs and there was a deficit of between 20-50% varying by age. Families in receipt of maximum benefit payments still needed an additional £30-£80 per week in order to meet their needs.

It is well known that many parents, particularly mothers, have no choice but to cease employment in order to care for their child full time at home. Employment opportunities are poor for mothers wishing to continue working and many barriers prevent them from returning to work. Steele and Davies (2006) report that in some families, parents continue to work at the early stages but as the child's condition progresses, they can no longer manage both roles and ceased employment. They also found that it was more common for mothers to stay at home. The families that were self-employed had to close their businesses and several families were in receipt of welfare payments. Families faced the double impact of losing their previous income and also the income that they could potentially earn in the future. In most cases, the parent who continued to work had to take on overtime to support the family. All families reported being less well off financially as a result. One study in the US, investigating reasons for reduced employment amongst mothers caring for a technology dependent child, found that single parent status, less available child care hours and poor family support were correlated with motives for

suspending employment (Thyen et al., 1999). Similar research conducted in the UK by Dobson and Middleton (1998a) found that even if parents desired to return to work, they were faced with the additional challenge of finding specialised child-care facilities, which can be costly and largely unavailable. Other obstacles highlighted include the potential loss of benefit payments and other forms of help such as home help and nursing care because they would exceed certain income thresholds.

2.12.1 The specific Costs of Caring

The specific examples of costs associated with caring for a child with a LLC, according to a large UK study by Hunt et al. (2013b) of (N=1,000) families, include nutrition and diet costs; for example, nutritional supplements like omega 3, high and low fat diets, boxed milk, steroid treatment which has the consequence of increased demand for higher food intake. Special clothing and equipment is another example of additional costs and the cost of cleaning clothes and bedding because of incontinence. Special footwear can cost up to £500,00. Other cost examples include transport and travel for regular appointments, heating and parking.

2.12.1.1 Cost-Effectiveness of Different Models of Care

Evidence on the cost-effectiveness of different models of care is lacking and more evidence is needed, so lobbying for suitable funding to meet the needs of parents and families can be done with greater authority (Goldman et al., 2012). A recent systematic review, examining the cost effectiveness of providing care closer to home (CCTH), which they define as any model of care that acted to prevent immediate inpatient admission and / or enable a reduced length of stay for children (up to the age of 18 years) with acute, chronic, complex or palliative care needs (Parker et al., 2013), found evidence that CCTH can reduce the burden and expenses for families and also decrease acute hospital admissions. Some savings were made to health service providers and society as a whole. Recommendations were made for routine data collection on outcomes and costs to apprise and further develop the CCTH model. They concluded that a significant research agenda still

remains and recommended a research focus on families in socially disadvantaged areas, as the evidence base for this group is particularly low.

An important concern is whether home based care transfers the financial burden onto families from the State, comparative to hospital care. One clinical trial in the UK presents an economic evaluation of a paediatric hospital at home versus traditional hospital inpatient care (Bagust et al., 2002). Children generally suffered from less severe conditions (such as breathing difficulties, diarrhoea/vomiting, and fever), so caution should be taken when interpreting the results. However, they found costs borne by families were reduced by an average of 44% for homecare patients. A study by Revill et al. (2013) examined the costs of providing care from both the statutory and voluntary care provider perspectives in Ireland and found that it is nine times more expensive to provide care in an acute hospital setting (€156,282) in comparison to a home setting (€16,267). Analysis also revealed that families face significantly less direct costs when caring for a child at home (€2,620) in comparison to caring for a child in hospital (€22,261). Such additional cost reductions include; travel, babysitting costs, loss of earnings, parking for long stay visits and hospital accommodation. Cost reductions were found in Frates Jr et al. (1985) study where home mechanical ventilation provided to technology dependent children cost on average \$10,000 dollars monthly in comparison to a registered nurse or hospital care that costs an average of \$16,000 a month.

It is clear from the research literature reviewed that families face considerable financial vulnerability when caring for a child with a disability. However, studies reviewing the costs associated with caring for a child with complex needs report wide variability in the methodology used, uniformity in data collection methods and a lack of standards on which to judge cost findings (Anderson et al., 2007, Parker et al., 2013). This reduces our ability to define the real magnitude of costs associated with caring for a child with LLC's. The present study examines out of pocket costs for families caring for children with severe disabilities in Ireland and attempts to assess and analyse the financial impact on families caring for children with life-limiting conditions in Ireland. There is a significant gap in the literature on the additional costs of caring, and this is addressed by this present study.

2.13 Scare Resources

Overall, Ireland has experienced decreases in the health care budget since the economic recession, resulting in a leaner public health service that was required to be more efficient and better integrated to deliver maximum value for money (Health Service Executive, 2013). Voluntary organisations also experienced challenges as less people were inclined to donate to charities and statutory funding was reduced from 10% to 8% which results in cut-backs to essential support services for children and young people, older people, people with disabilities, carers and rural communities. Decisions about the allocation of resources are constrained by poor funding and the availability of tools to aid decision-making. One palliative care trust for children with life-limiting conditions in England, “Jessie May”, has implemented an innovative tool; a Framework for Respite in Partnership with Parents (FRiPP) to enable a standardized and equitable allocation of limited resources (Bowman, 2011). Families score themselves on perceived needs and the trust informs them of available resources and a care agreement is compiled and a service offer made. They found that having a partnership model rather than a consumer led model helps to manage parents’ expectations. This model encourages the active participation of service users and moves away from outdated professional led models of telling families what they need. Parents reported satisfactory outcomes with the transparency and improved information sharing of the FRiPP tool. The quantitative element of this present study utilizes a novel way to elicit parents’ preferences of care packages by presenting various hypothetically designed packages of care, which when selected by parents reveals latent preferences which can be used to design more cost effective packages of care. This stated preference method is discussed in more detail in Chapter 6.

2.14 Summary

This literature was reviewed in relation to children with life-limiting conditions and is presented in three main sections: 1) the nature and experiences of caring for a child with a life-limiting disability and 2) the impact on families and the coping strategies and resources used to ameliorate their situation and 3) reviewed the financial costs of caring. Many of the studies that exist are limited due to sample

size, a lack of clear inclusion criteria, and generalizability of results. This is because of differing cultural norms and the heterogeneous health care systems reviewed. In the research literature, children with life-limiting neurodevelopmental conditions are included in many broad studies, which encompasses children with chronic illnesses, life-threatening and childhood disability. While some experiences and care needs are similar, unique differences still remain however, due to the differences in diagnosis, care trajectories, complexities of their illnesses and the terminology used to define them. This makes it challenging to apply the findings from this literature review that is applicable to the children in this current study.

There is a large body of research relating to families experiences of caring but there is a lack of evidence available that specifically examines carers' service experiences. Qualitative studies form a large proportion of the evidence out there on care experiences and quantitative studies are lacking. There is no evidence of studies using quantitative methods to inform the allocation of resources or preferences of care services. Cost studies are emerging but in most cases are dated. There is a large evidence gap on the out of pocket costs of caring for a child with complex disabilities and the impact it has on the family. It is the aim of this research to address the gaps identified and to define the experiences of using services, preferences of care and the financial impact of caring for families caring for children with life-limiting illness in Ireland.

Chapter 3

Research Design

3.1 Introduction

The purpose of this chapter is to provide an overview of the current study, along with the methodological approach and design used. The chapter commences with an outline of the aims and objectives of the study, the target population, participant criteria and the sampling methodology used. A discussion on the mixed methods design follows, along with a description of the ethics approval process and the ethical principles that were adhered to throughout the study. A more detailed outline of the methodologies, used for each of the respective study phases, is presented in chapters four to six.

3.2 Aims and Objectives of the Study

The study progresses in two phases; the first examines parents' lived experiences of caring for a child with a life-limiting neurodevelopmental condition at home. This involves gathering the perspectives of families using a qualitative technique. This had three main objectives:

Phase One:

- (1) To explore parents' lived experiences of caring for a child with a life-limiting condition at home
 - (2) To explore parents' experiences of using care services on behalf of their child
 - (3) To examine parents' perceived emotional and financial challenges of providing care
-

Phase Two:

The second phase of this study involves a quantitative technique, where families complete questionnaires that revealed their preferences for health care services to meet the needs of their child. This had the following two aims:

- (1) To elicit parents' specific preferences of care services and to estimate parents valuation of services
 - (2) To describe the additional financial costs associated with caring for children with LLC's living at home in Ireland
-

The specific research questions to be addressed are presented in the respective chapters relating to phase one and two.

3.3 Target Population

The target population is parents of children who have life-limiting neurodevelopmental conditions and who are not expected to live to adulthood. These children have a varied range of neurodevelopmental diagnoses and have extensive experience of health care services in the community. Participants were recruited through a voluntary organisation, the Jack & Jill Children's Foundation (JJF). The foundation is a registered Irish children's charity (Reg. number: 12405), which provides specialized home nursing care to families of children born with life-limiting neurodevelopmental conditions. The foundation has provided nationwide care across the 26 counties of the Republic of Ireland for the last 16 years. The foundation primarily relies on voluntary contributions and fundraising, and in addition to this, the Irish government provides 30% of their overall annual costs of three million euro.

The focus on this organisation arose because families receiving care from this organization have extensive experience with a myriad of care services in the community. Furthermore, this population was selected because they have a more comprehensive knowledge and experience of care services in comparison to

families based in an acute hospital setting. Families based in acute hospital settings are difficult to contact, have little to no experience of services in the community and are usually at a very sensitive stage of their child's illness trajectory. Therefore, the service users of the voluntary organisation served as the best possible target population for which to conduct this study.

3.4 Participant Criteria

The researchers sought the expert opinion of the nurses at the voluntary organization to inform inclusion and exclusion criteria. After explicit criteria were agreed upon, participants were identified through the voluntary organization database. Participant criteria is outlined in **Table 3.1** below:

Table 3.1 Participant Criteria

Participant Criteria

Inclusion criteria:

- Children with a diagnosis of a life-limiting neurodevelopmental condition.
- Parent (or) parents providing full time care at home.
- Parents caring for children aged between 1-7 years.
- Parent participants were required to be English speaking.

Exclusion criteria:

- Families newly recruited to the foundation and at early stage of their child's illness.
 - Families whose children were at advanced stage of illness.
 - Children under the age of one year.
-

Children under the age of one year were considered to be at a very sensitive stage of their illness trajectory and so it was agreed to set an age limit of one to protect the families in this study.

3.4.1 Sampling Procedure

In Ireland, it is estimated that there are approximately 1,400 children living with life-limiting conditions (Laura Lynn, 2013), however, reliable estimates are difficult to derive and this number encompasses a broad range of diagnoses, with varying degrees of severity. According to Collingridge and Gantt (2008), the sampling procedure used should relate specifically to the study rationale and specific research purpose. The purpose of this study is to provide a detailed evidence base for the development of care services that relate to children with life-limiting neurodevelopmental conditions at home in Ireland. The Jack and Jill voluntary organization finances nursing care for this population of children at home and has an average annual referral rate of 150 families. The JJF represents up to 85% of all children born with life-limiting neurodevelopmental conditions in Ireland and was therefore the most appropriate organisation from which to recruit. Sampling methodology and sample size justification is a contentious issue in qualitative research, with methods and the rationale for data saturation being poorly reported (Marshall et al., 2013); in particular, the quality of sampling procedures in end of life care research has been found to be poorly justified, with studies only focusing on one specific health care setting (George, 2002). Limited guidelines are available in relation to sample size requirements for data saturation and specifically in relation to palliative care research.

Common methods used in qualitative sampling include: convenience, theoretical, snowballing and purposeful sampling. The most appropriate method to use for this study was a purposeful (non-probability) sampling method, which “intentionally recruits participants who have experience with the central phenomenon being explored in the study” (Creswell, 2013 p.173-174). Two hundred and eighty families were registered with the foundation at the time of recruitment in 2012, as presented in the sampling frame in **Figure 3.1**. After exclusion criteria were implemented (see Table 3.1), 97 families were excluded from the study, leaving 184 families to contact for recruitment. Initially, 37 families responded, so in order to encourage a higher response rate, contact was made with the organization and nurses (gatekeepers) were asked to encourage eligible families to participate. This was a gentle process where families were reminded that they had received documentation about the study. Due to their hectic schedules, many

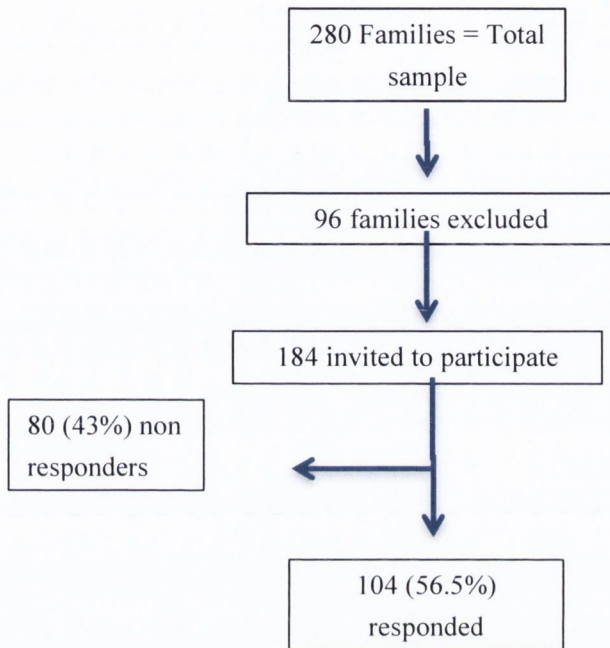
families said they did not have time to read the forms. In all cases, families were permitted more time and subsequently returned their signed consent forms, representing their willingness to participate.

The final response rate was 56.5%, with a total sample of 104 families willing to participate. Sample size is determined by the optimum number required to allow a valid inference to be made about the population (Marshall, 1996) and the final response of 104 families was considered to be strong, given it is recommended that a minimum of thirty-five to fifty is suitable (Sandelowski, 1995). Also, Irish research studies in the area of paediatric palliative care utilised similar sample sizes (Redmond and Richardson, 2003, Nicholl and Begley, 2012).

As this is a mixed methods study with two separate phases, two sampling procedures were used. The qualitative study involved conducting interviews with families, where 20% of the overall sample (N=104) were selected to participate in the interviews (24/104). This was in line with recent recommendations to include between twenty to thirty participants for interviews (Marshall et al., 2013). The second sampling procedure for the quantitative phase is more detailed and is discussed in Chapter 6. Finally, the non-response rate was 43%, which we considered quite low.

3.5 The sampling frame

Figure 3.1 Sampling Frame



3.6 Data Collection

A semi-structured interview technique was used in contrast to structured and unstructured interviews, as they are a more restrictive method of data collection for the purpose of this study. Structured interviews are less flexible and were not suitable for the chosen methodology, namely, interpretative phenomenological analysis (IPA) (discussed in more detail in chapter 4). Additionally, unstructured interviews would not be appropriate given that an interview schedule is typically used in IPA research. Semi-structured interviews are adaptive and allow one to probe further into areas of interest and were the most appropriate method to use for data collection in phase one of this study. Data was collected from parents across the Republic of Ireland using semi-structured in-depth interviews (n=24) and subsequent quantitative questionnaires (n=69).

3.7 Mixed Method Design

Mixed methodology is defined by Johnson and Onwuegbuzie (2004 p.17) as “the class of research where the researcher mixes or combines quantitative and qualitative research techniques, methods, approaches, concepts or language into a single study” and essentially has been described as the “third wave” or third research movement. Adopting a mixed methods design helps to legitimatise the use of both the qualitative and quantitative approaches thus fulfilling the overall objectives of this study which required the use of both. Using both methodologies neutralises the potential bias that comes from using only one method (Creswell, 2013,P.15) and provides a more comprehensive understanding of the topic under investigation (Creswell, 2013 p.4).

An important consideration in mixed methods research is that both qualitative and quantitative methods have significantly different approaches to analysis and interpretation of research. Interpretative phenomenology has a uniquely different set of principles in contrast to that of stated preference theory. Without acknowledging that each method explores the phenomena differently, we fail to recognise the unique contribution each approach can add to our understanding (Bryers and Pitchforth, 2014). One of the challenges of mixed methods research is dividing limited time among the different methods employed and consequently running each one to a subsidiary level. A strength of this study is that the same sample was used for both the qualitative and quantitative elements which allowed more time for concentrating on the quality of the methods used. Pragmatic time frames combined with a research agenda helped to abbreviate the process of the qualitative research as suggested by Peck and Secker (1999). Using IPA in a mixed methods study is uncommon and Smith (2011) has called for IPA to be used more in mixed methods, particularly in health research, because it lends itself well and can take a number of different forms. He further cautioned that there is a need to give due weight to the contribution of the qualitative arm. Typically, in IPA research, a small number of interviews are conducted but in this present study, the number of interviews were expanded in the hope to balance the contribution of both the qualitative and quantitative arms of the study. The overall goal of the mixed methodology was to draw from the strengths and minimise the weaknesses of both the qualitative and quantitative methodology. Mixed methods is supposed to be

expansive, inclusive, pluralistic and complementary (Johnson and Onwuegbuzie, 2004). According to Creswell (2013), mixed methods designs have a pragmatic worldview, in that the researcher bases the enquiry on the assumption that collecting heterogeneous types of data facilitates a more thorough understanding of a research problem that either quantitative or qualitative could not attain alone. This was the goal of this present study. Tashakkori and Teddlie (1998) outlines three distinct mixed methods designs:

- **Sequential studies:** The qualitative phase is conducted first and the quantitative phase then follows. Both phases are conducted separately.
- **Parallel / Simultaneous studies:** Both the qualitative and quantitative research is conducted in unison.
- **Equivalent status designs:** Both the qualitative and quantitative methodologies are used about equally to understand the phenomenon.

A sequential mixed methods design was used in this study which has two distinct designs; explanatory and exploratory. The *explanatory* design is where initially the quantitative data is collected and analysed and subsequently, qualitative data is collected and used to assist in explaining and interpreting the quantitative findings. It is explanatory because the initial quantitative results are explained further by the qualitative data. In contrast, the *exploratory* design is when qualitative data collection occurs first and is used to explore the views of participants. The data is then analysed and the findings are used to build the quantitative phase. The qualitative phase may be used to build the quantitative instrument which was a primary goal of this research. As a result, the exploratory mixed method design was chosen for the present study. The findings of the qualitative and quantitative phases are then integrated during the interpretation phase (Creswell, 2013 p.24).

3.8 Ethical Considerations in the Design of the Study

According to Grafanaki (1996), the three fundamental principles of ethics that generate serious challenges for qualitative researchers are: 1.) informed consent, 2.) confidentiality, and 3.) prevention of harm. Caregivers of the chronically ill (as is

the case for this study) are considered vulnerable populations in the context of social research (Liamputtong, 2006) and as this study involves investigating sensitive topics with families caring for a child with a LLC, it therefore falls within the sensitive research category. Sensitive research is defined as “research which potentially poses a substantial threat to those who are, or have been, involved” (Lee, 1993 p.4). Vulnerable participants find themselves at a higher risk of exploitation and the likelihood of maltreatment through research (Schrems, 2014). Protection of participants started at the early stages of the research design, where we sought to decrease research related vulnerability through identifying potential repercussions of the research (Creswell, 2013 p.77). Strong ethical principles involve “implicitly recognizing, acknowledging and affirming the dignity of our participants” (Seidman, 2012 p.139).

According to Kvale and Brinkmann (2009 p.63) ethical issues need to be considered at seven individual stages of the research process; these are highlighted in **Table 3.2** below. These were adopted as a strategy to safeguard participants from harm throughout the duration of the study.

Table 3.2 Ethical Issues at Seven Stages of Research (Kvale & Brinkmann 2009)

1. *Thematising*: The value of knowledge sought should be considered in line with potential improvements to study participants
 2. *Designing*: Consideration of core ethical issues, informed consent, confidentiality and consequences of the study
 3. *Interview situation*: Consideration of the personal consequences of the interview interactions i.e. stress and changes in self-understanding
 4. *Transcription*: Confidentiality of the interviewees’ disclosures needs to be protected to ensure the transcribed text is loyal to oral statements made
 5. *Analysis*: Consider how deeply the data will be analyzed and if participants will have a say on how statements are interpreted
 6. *Verification*: Verify knowledge represented
 7. *Reporting*: Consider the consequences of published work for interviewees’ and groups they belong to
-

3.9 Ethics Approval

To begin the approval process, a meeting was held with the Chief Executive Officer and his team from the Voluntary organization to discuss the research project and study objectives. A proposal was drafted and ethical approval was sought (Sep 2011) and granted (Jan 2012) by the Department of Health Policy & Management / Centre for Global Health Research Ethics Committee, Trinity College Dublin (Appendix 2). In addition, ethics approval was sought and granted by the voluntary organization to conduct the study (Appendix 3).

3.10 Informed consent of Participants

Informed consent is a tool and an operationalized form of self-determination to support potential participants to decide if they should or should not agree to participate in the study (Schrems, 2014). Before making the decision to participate in the study, an initial letter (Appendix 4) was sent from the voluntary organization to families within the inclusion criteria to inform them of the study. Following this, a letter of invitation (Appendix 5) was sent to families from the researcher inviting them to participate in the study. In addition, a comprehensive participant information leaflet was enclosed (Appendix 6), which described in detail the nature of the study, the research objectives, what would be expected of participants; as, “volunteering to participate in a research project should be based on being informed, not persuaded” (Seidman, 2012 p.140).

Details on data management procedures were provided, along with the contact details of the researcher. Parents were encouraged to contact the researcher with any questions they would like answered. A consent form (Appendix 7) was also enclosed for parents to sign, to signify informed consent to participate. A time frame of ten days was given to consider participation. A stamped addressed envelope was provided to return signed consent forms to the voluntary organization. Positive responses were compiled and sent to the researcher. Informed consent was treated as an on-going process and not a one-off event throughout this study, as

recommended by Richards and Schwartz (2002) and DiCicco-Bloom and Crabtree (2006). On-going consent is where parents were asked at various stages throughout the duration of the study if they still wanted to participate. If at any point, parents found it stressful, they were informed that they could discontinue participating at any time without consequence.

From the positive responses (N=104), twenty-four families were subsequently selected to participate in a semi-structured interview. Families were contacted by telephone and the researcher formally introduced themselves and reminded participants of the purpose of the study. This time was used to answer any questions and discuss potential interview times and locations. This process is discussed in more detail in chapter 4. Verbal and written informed consent was obtained from each respondent in the study.

3.10.1 Autonomy in Selecting Interview Location

Families were provided with the autonomy to select interview locations. All families requested that the interview take place at their home, with the exception of one parent who wanted the interview to take place at a location near a hospital appointment. Following this request, a letter was sent to parents requesting permission to use their home as the interview location (Appendix 8). All forms were signed and returned by stamped addressed envelope to the Department of Health Policy and Management, Trinity College Dublin. Conducting interviews in the participant's home is known to reduce potential hierarchy between the researcher and the interviewees (DiCicco-Bloom and Crabtree, 2006). In addition, it can help parents maximize control in their home setting (Coad et al., 2014).

3.11 Confidentiality of Participants

At the outset of the study, parents were informed that the information obtained about them would be used only by the researchers involved in the study and for the particular reasons outlined in the information leaflet (Appendix 6). Israel and Hay (2006 p.10) point to that fact that “researchers should be faithful to the obligations relating to respect for autonomy, justice and utility that are imposed by their

relationship with participants”. To protect the confidentiality of parents, children and families, the following measures were taken to ensure participant anonymization; during the data analysis phase, some passages of text were removed prior to analysis, which included, geographic location, children’s identities (names, age and diagnosis) and parents’ identities (name and occupation). We also paid special attention to health care personnel identities and removed any potential identifiers. A unique identifier code was assigned to each participant in the study and upon commencing the write up, the data was screened for vernacular terms which were removed from the text to ensure participants were not easily identifiable

Although careful measures were taken to protect the identity of participants, it is possible that participants could still be identified through speech characteristics and contextual features, which presents a challenge to researchers on predicting what will lead to association (Richards and Schwartz, 2002). In order to maximize the protection of participant’s identity, a researcher familiar with this population group reviewed three transcripts to screen for identification factors; anything that was identified as a concern was removed. Data remained on a password-protected laptop at all stages throughout the duration of the study.

3.12 Beneficence and Non-maleficence

The principle of beneficence is to provide benefits to others; its most simple form is altruism, love and charity, a responsibility not to inflict harm on others (Beauchamp and Childress, 2001). As this study involves parents describing experiences of caring for a child with a LLC, it was likely that emotional topics would be discussed, which has the potential to cause psychological distress (Addington-Hall, 2002). If participants became distressed during the interview process they were gently informed that they could take a break or discontinue the interview, as recommended by Orb et al. (2001).

Parents were informed of the potential benefits of participating in the study, both at the recruitment, phase and upon commencing all interviews. The usefulness of sharing their experiences and its potential to increase awareness of the needs of children with LLC’s were outlined. Many parents were keen to speak about their

existing experiences of care service in the hope of providing support to other parents or by affecting policy on a wider level. In addition, most parents expressed that they rarely talk about “their story” and felt it provided catharsis.

The researcher kept in mind that openness and intimacy can be characteristic of qualitative research, which can lead participants to disclose information that they may later regret (Kvale and Brinkmann, 2009 p.73). An example given by Rennie (1994) to reduce the occurrence of this would be to have parents listen back to and provide commentary on the recording to ensure they were happy with what was disclosed; this, however, was not feasible in our case. Therefore, all parents were asked if they were comfortable with what was disclosed at the end of each of their respective interviews and continued consent was obtained. The concepts of beneficence and non-maleficence are interlinked and if possible should be balanced in equal proportion (Beauchamp and Childress, 2001).

3.13 Conclusion

Chapter two outlined and discussed the aims and objectives of the study, the choice of mixed methods design and the ethical principles adhered to in the study. Additional discussions on the ethical principles adhered to throughout the study are highlighted, where relevant, in the subsequent chapters.

Chapter 4

An Examination of Parents Experiences of Using Services on Behalf of Their Child: An Interpretative Phenomenological Analysis

4.1 Introduction

Utilising care services is a salient feature of the lives of children with life-limiting conditions, where hospital doctor appointments can range from 1 to 104 visits annually (Sloper and Turner, 1992). In Ireland, there are many independent voluntary and statutory care agencies providing care to children with LLC's in the community, which presents additional challenges for parents endeavouring to procure the necessary support services. Care in the community for children with LLC's has been described as a fragmented model of care (Hunt et al., 2013b), with many independent agencies involved in the provision of care. Understanding parents' experiences of utilising care services on behalf of their child can help us to develop services in a way that parents really value. As highlighted in the literature review in Chapter 2, previous research in Ireland has focused on parents' experiences of caregiving (Nicholl and Begley, 2012, Redmond and Richardson, 2003), while other UK based studies have focused on experiences with specific diagnoses (Maxted et al., 2014, Glasscoe and Smith, 2011) and on adult children with neurological conditions (Kola et al., 2014). While these studies add significant and important insights into the nature of caring, this study adds a unique contribution in that it specifically focuses on parents' lived experiences of using services on behalf of their child. It is hoped that this research will contribute to the significant research agenda outlined by Emond and Eaton (2004). This research can be used to inform and develop care services for children with life-limiting conditions that are being cared for at home.

The purpose of this chapter is to describe the process of conducting the qualitative study. The chapter commences with a description of the research context and is followed by a description of the methodology used. The steps involved in

data collection and analysis is presented and the chapter concludes with a discussion of the strengths and limitations of the study.

4.2 Research Objectives:

The primary focus of this study is to examine parents' experiences of caring for and using services on behalf of their children with life-limiting neurodevelopmental disabilities at home. Considering the choice of methodology and the wide-ranging investigation the study sought to undertake, the broad research objective encompasses a number of specific research aims which are outlined below:

Table 4.1 Research Objectives

-
1. To examine the lived experiences of parents caring for and using services on behalf of their child with LLC's at home.
 2. To elucidate parent's perceptions and views on health care services for their child.
 3. To obtain a description of the services parents use to inform the quantitative part of this study (for stage two of study, referred to in Chapter 2).
-

4.3 The Parents and Children in the Study

The parents in this study are caring for children that were born with, or have developed, a life-limiting neurodevelopmental condition; all of which, have a broad range of neurological diagnoses and in most cases are technology dependent. Technology dependent children are defined as 'having dependence on a technological device to sustain life or optimise health and have a need for considerable and complex nursing care for substantial parts of the day or night' (Glendinning et al., 1999 p.35). The children in this study are categorized as having no reasonable hope of a cure, therefore, the goal is to provide the best possible quality of life for the child. Parents have extended roles, in that they are parents that

assume the role of a carer and also perform medicalised nursing activities. All of the families in this study are in receipt of additional support provided by the Jack & Jill Children's Foundation (JJF) and in, most cases, receive support from other voluntary and statutory agencies. The JJF provides families with a nursing care budget and a liaison nurse (key-worker) service. The liaison nurses remain in close contact with families and provides information on the additional supports available to them.

Parents have direct and intimate experience of providing care to their children at home. Providing care for children is demanding, requiring 24-hour monitoring and attention. On a daily basis, parents engage with health care service providers and maintain a busy appointment schedule for their child. In navigating the system, and obtaining services for their child, parents have unique and valuable insights into the management of care for children with LLC's in the community. Engaging with parents, to elicit their experiences and views, provides important and valuable insights into the development of care services for children with LLC's.

4.4 Methodology Orientation

The methodology chosen for this study is Interpretative Phenomenology Analysis (IPA), which seeks to explore an individual's personal account of an event, and in doing so, describe the lived experiences in order to elucidate the meaning that these experiences have for individuals (Smith, 2010). The aim of IPA is to explore in detail how participants are making sense of their personal and social world; the main currency for an IPA study is what the meanings, particular experiences, events, or states, hold for the participants (Smith et al., 2003). This study involves a number of in-depth semi-structured interviews with parents caring for children with a life-limiting neurodevelopmental condition at home in Ireland.

4.4.1 IPA and its Philosophical Underpinnings

IPA has its origins in phenomenology and symbolic interactionism, which, as defined by (Brocki and Wearden, 2006 p.88), holds that "human beings are not passive perceivers of an objective reality, but rather that they come to interpret and

understand their world by formulating their own biographical stories into a form that makes sense to them". IPA is committed to an emphasis on the individual, where participants are located in their specific contexts, and their experiences are explored from a personal perspective (Smith, 2010). IPA is phenomenological because it seeks to explore the experience in its own terms and not from predefined category systems (Smith et al., 2009). IPA's uniqueness is best captured in terms of the approach it offers, rather than a particular set of 'analytic steps' (Brocki and Wearden, 2006) where the phenomenological element captures a rich description of a particular event or experience, and the interpretative element considers the meaning of such rich descriptions (Smith, 2010).

The framework on which this study is based is grounded in the epistemological and theoretical, a position which is adopted within this study, through the lens of the interpretivist paradigm. The interpretivist paradigm is "informed by a concern to understand the world as it is, to understand the fundamental nature of the social world at the level of subjective experience" (Burrell and Morgan, 1979 p.28). According to O'Donoghue (2006 p.17) interpretivism has four underlying principles,:

1. If we want to understand a certain phenomenon, we must engage with every day activity to fully comprehend it.
2. There is always some autonomy and freedom in everyday activities.
3. Our interpretation of the phenomenon is influenced by what we currently think we know about it.
4. In everyday activity, we modify our understandings and meanings through the process of negotiation.

Constructionism is the theoretical framework that guided the qualitative study, and fits with the interpretivist paradigm in that "knowledge is established through the meanings attached to the phenomena studied and that researchers interact with the subjects of study to obtain data" (Krauss, 2005 p.759). My interest in the area of paediatric palliative care stems from a baseline study I was involved in, which examined the costs and outcomes of alternative models of care for children with complex life-limiting conditions in Ireland (Reville et al., 2013). At the outset of this current study, I was interested in eliciting parent's preferences of care

services using a quantitative experiment. However, due to a lack of evidence, I felt it was important to engage with the families beforehand, to examine parent's everyday experiences of using services on behalf of their child as I was not a clinical practitioner and had limited knowledge of the field. The interpretivist stance appealed to me, as it involved the researcher engaging with the parent's world to try to fully understand the phenomenon of caring for a child with a life-limiting condition.

4.5 Methodology

4.5.1 In-depth Interviews

The purpose of the in-depth interview is not to test a given hypothesis or conduct an evaluation, it is to understand the participant's experiences and the meaning they make of that experience (Seidman, 2012). In-depth interviews are seen as beneficial in comparison to survey methods, which are deemed an unsuitable way of investigating aspects of experience. According to Lee (1993 p.104), "Interviews provide a means of getting beyond surface appearances and permit greater sensitivity to the meaning contexts surrounding informant utterances." Legard et al. (2003) outlines three key features of In-depth interviews. Firstly, the topic guide used during in an in-depth interview is sufficiently flexible to allow for topics to be further explored. It also permits responses to be probed further if necessary which encourages more spontaneity. Secondly, in-depth interviews are interactive in nature and the material is generated by asking questions in a way that encourages the interviewee to talk freely when reciprocating. The third feature is that a range of probes and techniques can be used to achieve in-depth responses that reveals the reasons, feelings, opinions and beliefs of the interviewee. In-depth interviews enable the researcher to move beyond surface level responses and, so, was particularly suitable for Interpretative Phenomenological Analysis.

4.5.1.1 Constructing the Semi-Structured Interview Protocol

Developing an interview protocol prior to interviewing participants allows the researcher to think about the areas to cover during the interview process. It also ensures careful consideration of the wording used for questions that might be

sensitive for the respondents to answer (Smith, 1995). Smith's 1995 guide for the interview protocol design was adopted for this study and includes the following considerations:

1. To determine the general issue to be investigated during the interview process, which for this study involved clarifying the parents' lived experiences of caring for a child with a life-limiting condition at home. Three wide-ranging areas to cover during the interview were identified: 1) Parents' initial experiences of care services both in the acute hospital setting and in the community, 2) parents' experiences of utilizing care services on behalf of their child in the community, and 3) parents' emotional and financial experiences of providing care to their child.
2. Smith (1995) suggests deciding on the most appropriate sequence for the questions, so they run in a logical order. Interviews commenced with a general open-ended question, which asked parents to recall their initial experiences with the health care service at the early stage of their child's diagnosis. This then lead into questions about their transition from hospital to home and the type of care services they were receiving in the community. The order of questions reflected typical events experienced by parents. Smith (1995) also suggested identifying the most sensitive questions and placing those at the later stage of the interview so that respondents would feel more comfortable to answer more sensitive questions.
3. Appropriate questions were devised for each of the four areas, which are referred to in step 1.
4. Some questions were likely to lead to more questions, so potential probes and follow up questions for the interviewer to ask were considered. The final list of questions were neutral and open, instead of specific and leading. For the researcher, it was important to have openness in their approach and to hear and give credence to the responses of families (Morse and Field, 1995).
5. The interview protocol was designed and informed by the research objectives outlined in **Section 4.2**.

4.5.2 The Interview Schedule

In Figure 4.2 below, the outline of the interview schedule (Appendix 9) is presented in chronological order and asked parents about the following: demographics and background, pathways to care and description of services. This was followed with additional questions on parents' emotional and financial challenges and support needs.

Figure 4.2 Interview schedule

**Interview
Schedule**

Demographics and background: *Background of child's story, age and sex of child, child's diagnosis, primary carer, employment status, geographic location & family size.*

Pathways to care (i): *Transitions from hospital to home, service set-up with the Jack & Jill Foundation (JJF) i.e., the home care model. Identification of key people involved in the care plan, budget allocated to family, satisfaction with services & experiences with provision of care.*

Pathways to care (ii): *Other services: A description of supplementary services provided by the Health Service Executive and other voluntary sectors in the community, role of community health workers in care plan.*

Description of services: *Frequency and utilisation of care services in the community and service needs.*

Challenges facing these families which includes; *emotional and financial challenges.*

4.5.2.1 Preparation for Interviews

In preparation for the interviews, parents were individually contacted by telephone to inform them of the approximate duration of interviews (1 hour). In addition, the

background of the researcher was clearly explained and the study objectives briefly outlined. During these conversations, parents expressed enthusiasm about participating and the importance of sharing their stories in the hope of improving services for their child and others. This is consistent with other research studies where participants expressed altruistic views about participating in research (Horstman et al., 2008). The time was used to answer any questions parents may have had and was used also, as an opportunity to build rapport. Parents were informed that their refusal to participate at any stage would not jeopardize their child's care (Richards and Schwartz, 2002, Coad et al., 2014). The Interview schedule was piloted with two additional researchers, so it was refined and perfected before conducting interviews, in order to ensure consistent engagement throughout the interview process, as suggested by (Smith et al., 2009).

4.5.3 The Interviews

Upon commencing interviews, families were reminded briefly of the study objectives and verbal consent to participate in the study was obtained. As a safety measure, an interview checklist (**Table 4.1**) was used at the beginning of the interview process to make certain the correct protocol was followed throughout, as recommended by Tong et al. (2007):

Table 4.3 Interview Checklist

Interview Checklist
<ul style="list-style-type: none">• Respondent has read the information sheet• The information sheet was discussed with the respondent• Confidentiality and data handling procedures were discussed• Consent form was signed before commencing the interview• Permission to record the interview was granted

Agreed consent was treated as an ongoing process throughout the duration of the study (As discussed in detail in Chapter 2). Upon the completion of all interviews, parents were asked if they were happy with what was disclosed during the interview. Many sensitive topics were discussed during the interview process, so it was imperative that parents were comfortable with what they had disclosed and were happy to continue participating (Orb et al., 2001). Parents were again gently reminded that they could exit the study at any time without consequence.

4.5.3.1 Data Collection and Data Management

Interviews took place over a 13-month period, from June 12th 2012 to July 2nd 2013. The interviews ranged in duration from 20 minutes to 1 hour and 40 minutes, with the average duration of interviews lasting 56 minutes. **Table 4.2**, below, presents the duration of twenty-four Interviews. All interviews were audio-recorded using the free downloadable digital audio recorder software, Audacity® 2.0. In addition, an Olympus Digital Voice Recorder (VN-7600PC) was used during the interview process. All recordings from the Olympus recording device were saved onto the researcher’s password protected laptop.

Table 4.4 Duration of Interviews

Interviews (N)	Interview Duration
1	65 min 43 sec
2	75 min 20 sec
3	55 min 47 sec
4	40 min 36 sec
5	30 min 45 sec
6	24 min 38 sec
7	73 min 0 sec

Interviews (N)	Interview Duration
8	33 min 42 sec
9	37 min 24 sec
10	61 min 52 sec
11	63 min 32 sec
12	85 min 30 sec
13	68 min 50 sec
14	59 min 26 sec
15	44 min 9 sec
16	44 min 9 sec
17	109 min 0 sec
18	71 min 54 sec
19	75 min 37 sec
20	35 min 13 sec
21	20 min 56 sec
22	62 min 24 sec
23	56 min 33 sec
24	58 min 30 sec

4.5.4 The Transcription Process

In IPA research, the transcription process involves the semantic record of the interview, that is, the words spoken by all present should be transcribed and notes added where relevant to represent non-verbal utterances (laughter) (Smith et al., 2009 p.74). Spaces were left between each turn in the conversation and wide margins for ease of coding. The researcher transcribed interviews verbatim over the

period of September 2013 - March 2014. Individual transcriptions ranged in length from 4 - 15 pages of single text and individual transcriptions took on average two days to complete.

4.5.5 Analysis of Interview Data

This study used Smith and Flowers et al (2009) four step guidelines for IPA analysis, as outlined below. The interpretative role of the researcher was strictly adhered to during the data analysis process and existing knowledge was suspended in an attempt to see the world “as it is” from the researcher’s perspective (Flowers et al., 1998 p.412). The following section outlines the four stages of analysis and the validity exercises that followed:

4.5.5.1 STEP 1 - Reading and Re-Reading

To enter into the participant’s world, active engagement with the data is necessary. The first step involves engaging fully with some of the original data by re-reading original transcripts and listening back to original recordings. At this early stage, Smith and Flowers et al. (2009) suggest bracketing out the most noticeable revelations for a period of time, thus, reflective notes were taken throughout this research phase and contradictions and paradoxes highlighted. This stage of analysis was deeply engaging and took a considerable amount of time (8 weeks) to complete. Smith and Flowers et al. (2009) encourage the deconstruction of data where the researcher avoids focusing upon simplistic readings, therefore, some paragraphs were extracted and each individual sentence was read carefully line-by-line.

4.5.5.2 STEP 2 - Initial Coding

This step examines the semantic content and language use on a very explanatory level (Smith et al., 2009). The aim here is to produce a systematic and thorough set of notes and statements on the data, using exploratory commentary; descriptive (describing what the participant said), linguistic (the language the participant used)

and conceptual (engaging with the findings on a more interrogative level) ideas (Smith et al., 2009). To begin, paper copies of the transcripts were printed and sentences of importance underlined with provisional notes entered onto the right-hand margin. The meanings which participant's placed on particular events were noted in order to push the analysis to a more interpretative level. Identifying shared experiences across a relatively large sample size (n=24) would not be cost-effective, so a focus was placed on identifying themes that were shared by all participants, as recommended by Smith et al. (1999).

4.5.5.3 STEP 3 - Developing Emergent Themes

The 24 transcripts were entered into NVIVO Version 10 software. This software is useful for the management and analysis of large volumes of data. The diagrammatic feature also provides an opportunity to view additional perspectives and interesting aspects of the data that would otherwise be missed if manually analysed. The main goal at this stage of the analysis was to develop emergent themes. Reducing large volumes of data, while ensuring to capture the interrelationships and patterns in the data, is crucial. The analysis of complete chunks of data followed and individual sub-themes were grouped under super-ordinate themes in NVIVO folders. The themes represented a range of understandings that were relevant to both the participant and the researcher.

4.5.5.4 STEP 4 - Identifying Connections across Emergent Themes

This next stage involves the charting and mapping of how the themes fit together. To begin, themes that were both parallel and comparable were placed together; this formed clusters of related themes (Smith et al., 2009). Within step four, the following measures were used to develop connections across emergent themes:

- 1) **Abstraction:** where similar themes are compiled and super-ordinate themes are developed.
- 2) **Subsumption:** (similar to abstraction) where an emergent theme itself acquires a super-ordinate status (examples of which are provided in the results section “fight for services” in Appendix 11).

- 3) **Polarization:** where contrast (also known as deviant cases) between emergent themes is recognized i.e., those that are the opposite to emerging themes (Pope et al., 2000).
- 4) **Contextualisation:** highlight themes that relate to key life-events.
- 5) **Numeration:** take note of the number of times a theme is stated. Nvivo's useful feature, to highlight the most frequently used words, was utilized here to visualise numeration (Appendix 12).

4.5.6 Inter-rater Agreement on Coding

Brocki and Wearden et al. (2006) provide a comprehensive discussion on the validity exercises utilized by qualitative researchers and argue that IPA is inevitably subjective and that it is unlikely for two researchers, who are reviewing the same data, to produce the same results. Acknowledging the interpretative nature of the results is important as they are only a version of the truth (Richards and Schwartz, 2002). A number of validity exercises were conducted in this study which was guided by Kvale and Brinkmann (2009) seven stages of research ethics as discussed previously in Chapter 3. The first validity exercise involved a presentation of the results to the voluntary organization nurses and company chief executive (CEO). This exercise was extremely useful and opened up important discussions around the issues of caring for children in the community. The nurses in particular were able to verify the findings from the study and provide clarity where needed. Secondly, an expert in the field of qualitative research reviewed three interview transcripts and confirmed the findings before the joint thematic framework materialized. Finally, an independent researcher read final quotes and themes and agreed on the findings presented. Families were not approached to confirm the research findings; this was deemed impractical due to the time commitment it would take and due to the fact that the parents in this study, led hectic life-styles and it would be onerous for them.

4.6 Findings

4.6.1 Introduction

The focus of enquiry was to examine parents' experiences of caring for and using services on behalf of their children with life-limiting neurodevelopmental conditions at home. In the following section, a brief description of the alterations made to the transcripts is provided. A description of the characteristics of families and children who participated in the interviews is outlined. The chapter then proceeds with a presentation on the findings from the parents' interviews and concludes with a brief discussion on the study findings and limitations.

4.6.2 Alteration to Transcripts

To protect the identity of all study participants, unique identification codes were applied to each participant in the study. All quotes transcribed and presented were taken from the raw data. Changes to the quotes only occurred when it was necessary to clarify or give context to the participants' experiences.

4.6.3 The Participants

All parents involved in the study were from the Republic of Ireland. Individual addresses are not reported in order to protect the participants' identify. A total of 23 mothers and 3 fathers participated in the study. Parents ranged in age from 35 - 48, with the average age being 40. Present at the interview were both parents (in two cases), just the father (in one case) and just the mother (in the remaining 20 cases). At the time of the initial data collection, 16 mothers and 2 fathers had ceased employment to care for their child on a full time basis. Two mothers continued to work full time and three mothers reduced their work hours to part-time. Interviews took place in a broad range of geographic locations, as outlined in **Table 4.3** below.

Table 4.5 Geographic Location of Interviews

Location (ROI)	Number of Interviews
Republic of Ireland	
Dublin	4
Co Kildare	3
Co Tipperary	2
Co Mayo	2
Co Waterford	2
Co Roscommon	3
Co Cork	3
Co Monaghan	3
Co Wicklow	2
Total	24

Table 4.6 Marital Status of Parents

Marital status of parents	
In a two parent relationship	23
In a one parent relationship	1
Total	24

The children in this study (presented in table 4.5) had a range of heterogeneous neurodevelopmental conditions and disorders which varied in developmental domains such as; motor function, communication and language, cognition and activities of daily living as defined by Racine et al. (2014). Five of the children in the study did not have a specific diagnosis and were defined as either, “developmental delay”, “suspected unknown genetic disorders” or simply “no diagnosis”. All of the children were cared for at home on a full time basis but faced frequent hospitalizations as a result of the uncertain nature of their conditions. Most of the children required assistance with feeding and used percutaneous gastrostomy (PEG) or nasogastric (NG) tubes. Children needed assistance with mobility, personal hygiene and bladder and bowel management. In addition, children required medications for symptoms associated with seizures, progressive deterioration and acute illness. Children ranged in age from 1-7 years as outlined in Table 10. Typically, the provision of financial aid towards nursing care is ceased by the JJF subsequent to children turning four years of age. In two cases however, the foundation continued to provide funded nursing care.

Table 4.7 Examples of medical diagnosis similar to children in this study

Types	Similar diagnosis
Neurological* disorders	Cerebral Palsy
	Hydrocephalus
	Lissencephaly
	Spina bifida
	Neurodegenerative disorders
Genetic disorders	Chromosomal deletion syndromes
	Complex Cardiac disorders
	West syndrome

Types	Similar diagnosis
No diagnosis	Developmental delay
	No known diagnosis
	Suspected unknown genetic disorder
	Undiagnosed metabolic disorders

* Two families had more than one child with a neurological diagnosis

Table 4.8 Age range of children in study

Age range	
1 - 4 years	22
4 - 7 years	2

4.6 Explication of Parents' Interviews

This section is concerned with highlighting and revealing parents' experiences of using health care services on behalf of their child. The section commences with an overview of the themes identified as presented in **Table 4.9**. This offers a framework for a more thorough discussion of the themes that follows. Following in-depth analysis of the Interviews, an example of which is provided in Appendix 11), eight essential themes emanated from the analysis:

1. Transition from hospital to home
2. The perpetual wait for services
3. The crucial role of liaison nurse
4. Parents as advocates
5. Physiotherapy – a highly valued service
6. The economic costs of caring
7. Emotional effects of caring
8. Future concerns

Sub themes that allowed the emergence of these essential themes are presented in **Table 4.9** and are illustrated throughout the text.

Table 4.9 Overarching Themes and Related Sub-Themes

Master Themes	<i>Transition from hospital to home</i>	<i>The Perpetual wait for services</i>	<i>The vital role of the liaison nurse</i>	<i>Parents as advocates</i>	<i>Physiotherapy - a highly valued service</i>	<i>The financial costs of caring</i>	<i>Emotional effects of caring</i>	<i>Future concerns</i>
Sub-themes	Going home, a vulnerable time	The long wait for service set-up	Service set-up	The battle with service providers	Provision shortages	Additional out of pocket costs	Everyday challenges	A tentative future of care provision
Sub-themes	Isolation The importance of early referral	Too little, too late	Lobbying Emotional support and advice	Administration workload	Differing views	The medical card	The need for emotional support services Fear of opening up the wound	

Master Themes	<i>Transition from hospital to home</i>	<i>The Perpetual wait for services</i>	<i>The vital role of the liaison nurse</i>	<i>Parents as advocates</i>	<i>Physiotherapy - a highly valued service</i>	<i>The financial costs of caring</i>	<i>Emotional effects of caring</i>	<i>Future concerns</i>
Sub-Themes	<p>Missed opportunities</p> <p>The need for centralised Information</p>							

4.6.1 Master Theme 1 - Transition from Hospital to Home

Transitioning from hospital to home emerged as an extremely strong theme from the interview transcripts and represents a significantly sensitive time for parents during their child's care trajectory. Parents used terms like "disconnected", "vulnerable", "overwhelmed" and "an emergency" to describe their feelings of leaving hospital. The broad super-ordinate theme is captured by five sub-themes that characterize parents' experiences in this particular context:

- 1) Going Home – a Vulnerable Time
- 2) Isolation
- 3) The Importance of Early Referral
- 4) Missed Opportunities
- 5) The Need for Centralised Information

Going Home - a Vulnerable Time

Parents articulated some of the challenges which they experienced when transitioning from hospital to home with their sick child. Anxiety prevailed, especially in relation to the lack of pre-arranged service supports. In addition, a concern over the proximity of family members to provide additional supports perpetuated their anxiety. One parent (P24) expressed injustice at the presupposition of having to provide care to their child without any additional supports.

"I didn't know how the hell I was going to bring this baby home, I didn't have any family around me nor did my husband...I mean it's hard looking after a baby that's well. I had a real problem with the fact I was expected to go home without any kind of support. It was a real emergency coming home, it was all down to us" [P24]

Another parent (P22) experienced vulnerability at the sudden complete reliance on others to inform them on how to proceed next with caring for their child. It was also evident that parents were afraid of having to suddenly assume a

medicalised role. Information provided on services was inconsistent and vague, as one parent (P9) expressed retrospective incredulity at the fact that they were sent home with a baby that was failing to feed adequately:

“Some services were offered to you and some weren’t. You’re very vulnerable, you completely rely on other people to tell you about what’s the next step. You are so dependent on people, like you never were before. When I look back and think of those first few weeks, it was such a shock to have to suddenly prepare medicines” [P22]

“It’s funny when you think back on it now, they sent me home with a baby that wasn’t really feeding. Looking back on it, I know it was wrong. He was aspirating on us all the time” [P9]

Parents invest considerable amounts of time into attending medical appointments on behalf of their child. One parent below (P5) attended 65 appointments in the first year of their child’s life, indicating a frantic schedule for families in these circumstances:

“The first year, (child’s name) had 65 appointments, between everything, physiotherapy, speech and language therapy and hospital appointments” [P5]

Isolation

Initially, when children are based in hospital, a team of health care professionals surround parents to provide a source of comfort and support. However, when parents are discharged from the hospital to home they can feel cut off which leads to feelings of isolation and abandonment where they are left to deal with everything alone, as reported by (P21 & P7). Additionally (P7) also commented on the discontinuities between the hospital and all other services.

“When you’re at home. You’re completely cut off” [P21]

“Like there’s a disconnect in the hospital between all the services and appointments but like when you come home, you’re alone” [P7]

The Importance of Early Referral

Two of the twenty-four parent’s interviewed (P14 and P23) provided accounts of the transition from hospital to home that appear to contradict other experiences discussed above. Both parents had singularly positive experiences that were contingent on the fact that they were connected with community services before returning home.

“Our transition from hospital to home was very positive, the first week I was home, I had someone call every day so it was great. Well JJF were pounced on from the Public Health Nurse and the CNN nurse and so they were very prompt and came out to us” [P14]

“What I found amazing was that when the JJF came in, they were trying to set us up for coming home so that when we did come home, we had lots of things in place. Like they provided loads of practical information even down to tax information, even tips about negotiating what to expect from the HSE homecare package.” [P23]

These unique contrasting experiences, also known as “deviant cases” (Mays and Pope, 2000) provide alternative explanations on service experiences when transitioning from hospital to home; although only two cases reported such positive experiences, there is something to be gained by highlighting the vital roles of the public health nurse and the clinical nurse specialist in referring parents to essential services. Parent’s continuous engagement and strong communication with the voluntary organization prior to and upon returning home eased the transition and

made it a very positive experience. The immediate service set up and resultant outcomes highlight the essential need for early referral to ensure a smooth transition.

Missed Opportunities

Families have extensive information needs, both at the beginning and throughout the duration of their child's illness trajectory. Information and advice on supports available is an essential prerequisite for a successful transition from hospital to home for families. Parents (P9 & P14) explain below how failure to receive both timely and adequate information can intensify an already stressful situation and result in missed opportunities to avail of essential supports. Informing parents on available resources in a timely manner avoids unnecessary stress and extended hospital stays.

“He needed oxygen at night; they were saying have you got a medical card? I was like no. They were like; you won't be able to pay for the oxygen at night. This was the day before I was going home. I started to get into a sweat and a panic. Over three months, all I wanted to do was bring him home for good, no one told me to apply for a medical card...” [P9]

“I firmly believe there could have been more of a push to get that paper work in while we were back in the hospital. We could only claim back to the time when we had applied for it. It's a great payment and not making people aware that they should apply for it is a bit of a drawback” [P14]

Two parents (P22 & P7) report below how it was only when their child was re-admitted to hospital, that their child's needs were recognised and they were subsequently provided with the necessary and correct care services. Families who are left without the necessary supports suffer the consequences, which include sleep deprivation and increased stress. Services in the community are inadequately

provided to families and the disconnect between hospital and the community is evident. Supporting and managing families care needs in the community averts these crises from occurring:

“When he turned 1, he went in to hospital for three months. That’s when they realized that he needed nurses. His package was then changed to all nurses and no carers” [P22]

“We were both really suffering with sleep deprivation. We couldn’t feed him and he was getting sick all the time. We finally ended up back in hospital with him. I broke down again and that’s when the social worker finally got us 10 hours a week to help us at home” [P7]

The Need for Centralised Information

Having one streamlined source of information on available supports was very important to parents. Both parents (P21) and (P18) expressed frustration at the lack of such a service and the inherent difficulties faced when navigating a vague non-priority based system:

“There needs to be a centralised person or information system for families in this situation. It’s very hard to know what you should be looking at, like it’s time consuming. Filling out the medical card form is so difficult” [P21]

“Chasing services, it’s so frustrating. It’s not clear-cut. They should make it clear, like what’s there and if there are people that have more needs, they should be looked after first. They should make it a priority-based system if resources are tight. It shouldn’t be who screams the loudest. It’s just not fair” [P18]

4.6.2 Master Theme 2 - The Perpetual Wait for Services

This theme relates to the length of time, and perpetuating factors, that lead to extensive delays in the provision of care services and supply of essential equipment.

Two sub-themes relevant to the superordinate theme are highlighted below:

- 1) The Long Wait for Service Set-up
- 2) Too Little Too Late

The Long Wait for Service Set-up

According to the voluntary organization, the average life expectancy of children with neurodevelopmental life-limiting conditions is generally in and around two years of age. In this short time, it is imperative that children are provided with the equipment needed to sustain the best quality of life possible, however, parents report significantly long waiting times of up to two years for services, as outlined by (P7 and P13) below:

“I was a year waiting on a buggy which is ridiculous. I’m now two years waiting on a chair” [P7]

“The equipment side of things was very slow, the physio was supposed to come up and assess (child’s name) for a chair or a buggy and it was the following July before we got anything. (Child’s name) was over a year old at that stage” [P13]

Parent (P3) describe the frustration in instances when the equipment finally arrives but their child has outgrown it or the personnel responsible for installing it lack the necessary skills:

“I mean by the time we receive any equipment he’s outgrown it. (Child’s name) in that chair for three years and we’re still fighting to get him a proper one. I can’t bathe him here” [P20]

“They keep saying no to everything and it’s tiring having to ask again and again. Then when you do get something, the people arrive to put it up and they don’t have a notion how to install it or what it’s even supposed to be used for” [P3]

Children with LLC’s, that are technology dependent, can require up to 22 pieces of equipment to manage their needs at home (Nicholl et al., 2013). A failure to provide families with the necessary equipment in a timely manner has negative outcomes for both the child and family. One mother (P1) pointed to how satisfying it was when her child received the correct equipment but relayed her frustration and anger at the time wasted in obtaining it:

“The minute he sat into that chair, he sat up straight, he had his arms out and we thought...oh my god...he should have that months ago! You know that is frustrating.” [P1]

In the case of general service set-up, be it nursing care or equipment, both families (P4 and P14) did not receive any form of support until their children reached the ages of 15 – 24 months old:

“We didn’t get anything until he was two years old” [P4]

“(Child’s name) was 15 months before she started receiving care” [P14]

The extensive waiting times signify incongruities in the communication between service providers and parents and what is also evident, is a lack of consistency in the provision of care extended to families. In some cases, the length of time families wait for services, surpasses their child’s life-expectancy. This constitutes a central concern in the provision of care to children with LLC’s.

Too Little Too Late

Parents expressed anger and frustration at the fact they were offered services at a time when they were no longer needed as much. One parent (P18) talked of being pushed to breaking point before the services were provided and another (P22) commented on how, without the voluntary organizations support, they had absolutely nothing:

“We didn’t need them as much as we had done. If they had only given me the hours when I really needed them so I just went for it then. It felt like pay back, for years, they made us get to breaking point” [P18]

“I had the health nurse on to me and she was saying she was going to write to the HSE and I sort of said to her, I needed the help when I came home last December and only for the JJF I had nothing, absolutely nothing” [P22]

Caring for a children with LLC’s is demanding because of the constant monitoring that is required. Not having adequate supports in place can lead to high levels of stress; one parent (P16) describes an unremitting state of fear and emotion before they finally received support, after which, they reported having time to themselves. Another parent (P20) explained that the voluntary organisation was surprised to see how much they were doing on their own when they were finally referred to them (P20). Managing on their own seems to be a pervasive and ubiquitous issue for parents caring for a child with life-limiting conditions:

“We were in the house, in a constant state of fear and emotion. When we got the hours from the JJF. We were able to get some time off to ourselves” [P16]

“We should have been referred to the JJF, I mean it was 8 months before we were referred on to them. They were surprised by the

amount I had been doing on my own by the time I got in touch with them” [P20]

4.6.3 Master Theme 3 - The Vital Role of the Liaison Nurse

A Liaison nurse (also known as a link-worker) works for the voluntary organization and has a number of roles in helping with the family. They assess and provide direct funding to families based on need. In addition, they advise and educate families on care needs for such things as feeding, suction and medications. They liaise with a multidisciplinary care teams to coordinate care for families and when necessary lobby for services on behalf of parents. The role of the liaison nurse emerged as an extremely strong theme from the interview data. Families talked about how they valued the relationship they had with their liaison nurse and discussed the many roles they fulfil such as;

- 1) Service Set-up
- 2) Lobbying
- 3) Emotional Support

Service Set-Up

One of the key roles of the liaison nurse is to organize the necessary support services for parents to return home with their child. This strong emphasis and commitment to making things happen was favoured in particular by parents, as (P19) deliberates below. Parents’ contrasting perceptions of hospital staffs’ views on their child, in comparison to those of voluntary organization, are striking and reveal that each had a differing ethos towards the child. Parent (P12) talks about the technical support one liaison nurse provided, which shows the extent to how involved they are on all aspects of care:

“There was never a plan to get my child home, until I met (liaison nurse name). She had a lot of contacts. I felt like they (hospital staff) had written my child off. Everything got better when (liaison nurse

name) came on board. I could text her or ring her. She got the approval for the home care package. She got 50 hours for a carer” [P19]

“He was discharged from (hospital name) by gravity feeding. My liaison nurse walked into my house and said. What are you doing? Have you not got a pump? She had one organized within two days. I didn’t know anything about pumps” [P12]

Lobbying

Liaison nurses provide a voice for parents in what can seem like a daunting task of having to face a team of health care professionals alone. One parent (P8) provides an example where the liaison nurse stepped in and made an appointment with the hospital team to negotiate for night nursing care:

“(Liaison nurse name) was fighting with the HSE to get a home package. She wanted to get me a nurse at night. We held a meeting with the hospital team” [P8]

Emotional Support and Advice

It was clear from the interviews that parents build many special connections with their liaison nurses. The liaison nurse becomes a regular fixture in the family unit and both parents (P21 and P15) demonstrate the broad role nurses have and the emotional support, in particular, they provide:

“My Liaison nurse organized the nurses for me and even got one of the nurses from the hospital that we already knew so that was a great support. My liaison nurse tells me about entitlements and gives suggestions and ideas on things we can get. I also like just talking to her if I’m having a crap week or a good week, you know yourself” [P21]

“I get most of my advice from (Liaison nurse name). You’re always afraid with other services... they always have their own agendas. They say they have children’s best interest at heart... but they are stuck with numbers and budgeting. It’s nice to know what they are on your side” [P15]

4.6.4 Master Theme 4 - Parents as Advocates

The fight for services was an issue expressed by mostly all of the families interviewed in this study. Some of the key themes that encapsulate this can be described as:

- 1) The Battle with Services Providers
- 2) Administration Workload

The Battle with Service Providers

Parents describe an austere atmosphere in their mission to obtain services. A constant battle ensues where service providers are repeatedly saying no and parents are left exhausted. One parent (P23) describes, “fighting” and “screaming” for services and another (P21) noted the feeling of unworthiness that was implied when they did receive services. The outcome of this constant battle amplifies the already stressful situation parents find themselves in as expressed by parent (P15):

“You learn after a few months that you have to start fighting and screaming for services. I mean not having to fight the system that’s geared to say no and not dealing with 6 different people to get a buggy” [P23]

“I asked about home help and I applied. I had this horrible woman come back to me and say that I can get it for 8 weeks and that’s it and she was like, now I’ll give you this little bit, but you can’t come

looking for it again. She made me feel like I wasn't entitled to it and I was lucky to have it" [P21]

"Battling the services is stressful, having to use services is stressful, you know getting them into the car etc. You know it's all hard. It's all-difficult" [P15]

One parent (P24) describes having to ruffle a few feathers before services were provided. A key feature of the parents' experiences of using state services is the divisive culture that permeates and an undercurrent of "us versus them".

"I had to ruffle a few feathers and get them annoyed and next thing I get this number for this services and it's free" [P24]

In times of austerity, where budget cuts prevail, parents are constantly reminded of the lack of funds available to them. This extremely challenging environment, to which parents find themselves, is counterintuitive to recent policy objectives on providing optimal care to children with LLC's based on need.

Administration Work Load

Parents describe the considerable amount of time they invest in administration work to procure services (P23) and to maintain service provision (P17). The continuous trend of long waiting times associated with accessing services is omnipresent, as one parent (P8) describes the duplicitous task of sending documents back and forth only to have them rejected or disqualified:

"Every night you're writing a letter to someone, it has to be done. The administration is a big workload. Like if you don't stop then you're grand. I'd say if you stopped you'd probably collapse" [P23]

"I set out an hour every day to make the calls. Some parents get so tired and I do, because you feel that you shouldn't have to be

fighting for this service. Then I think it's best to keep going because what will happen to her if I let it get to me" [P17]

"I'm trying to get my rebate and get exempt from paying tax. I sent them all the necessary forms and they sent them all back saying it was wrong. Then I sent all the correct stuff back and then I got a letter saying the car had to be in my name... all these silly things keep happening, It's so difficult to get this sorted" [P8]

One parent (P21) below demonstrates strong organization skills, requiring nurses to fill out their prior availability in a notebook. Parents are required to attend many appointments, so a pragmatic approach allows them to plan ahead effectively. Parent (P21) is a self-starter and someone who likes to organize themselves, as opposed to leaving it up to someone else to dictate their schedule. What is also evident here is that parents are actively making the best use of the resources available to them:

"It is time consuming, I get the nurses to fill in notebooks, you know when they are going to be available a few weeks in advance so I can plan appointments around them, I prefer to have them come in the mornings so I make sure to use my budget for that" [P21]

4.6.5 Master Theme 5 - Physiotherapy: A Highly Valued Service

Out of all of the services discussed, the one that parents had the most difficulty accessing was physiotherapy. Physiotherapy is provided to children to help improve muscle strength, maintain muscle length and improve movement in their joints. Some parents receive this service at home, while others travel to utilize it; all of this depends on the family's geographic location and the flexibility of the care provider. The frequency of physiotherapist appointments varies from 1-4 hours monthly but

1-2 hours is typical. Appointments last 30 minutes to 1 hour in duration. The two key issues in relation to physiotherapy raised by parents was:

- 1) Provision Shortages
- 2) Differing Views

Provision Shortages

Parents expressed concern over staff shortages (P8) and in turn expressed apprehensions (P24 & P21) over having to perform the role of a physiotherapist themselves. They were concerned that they lacked the required knowledge to recognize if what they were performing was helping their child. A mother (P21) points out that her child can only endure physiotherapy for a few minutes at a time due to fatigue. Perhaps providing one hour long appointments, or in some cases 30 minutes, is too much for this patient group. Assuming the role of physiotherapist is distressing for parents in addition to the multitude of other tasks they have to perform.

“The physiotherapist and occupational therapist have just gone on annual leave and were never replaced. It’s hard because he’s not putting weight on his legs” [P8]

“She doesn't get as much to reach her full potential, her hamstrings are tight and they need to be worked on every day. She gets tired, so I have to do it for 2-3 minutes and give her a break, I mean you can't just do a 1 hour physio session... it's not like that. I need more of that kind of help; more frequency is what I really need” [P21]

Differing Views

Parents expressed a contrary view to physiotherapists in relation to their child's required needs. The disparity of views between parents and health care professionals is immediately evident from parents (P16 & P11) excerpts below:

“I’d love to get physio for her every single day...but it’s just not there. Like I don’t know, the Physio tells you that she doesn’t need it but I feel she does” [P16]

“I would like more Physio for (child’s name). Now that he’s getting a bit older, it makes a difference you know. The Physio guy tells me he’s getting all he needs but I feel my child needs more” [P11]

4.6.6 Master Theme 6 - The Financial Costs of Caring

In twenty-two out of the twenty-four households interviewed, one of the parents ceased employment to care for their child full time at home. This automatically reduced household income and placed families in precarious financial situations (Shearn and Todd, 2000). Parents consistently mentioned the additional costs associated with caring for their child at home. The overarching sub-themes that emerged under financial costs include issues around:

- 1) Additional Out of Pocket Costs
- 2) The Medical Card

Additional Out of Pocket Costs

Parents have no choice but to manage money as effectively as they can by allocating funds to specific items of importance, as one mother explained (P17); another parent installed a meter box (P7) to ensure electricity bills don’t escalate too high. Electricity bills are a consistent cost concern for parents and are a result of equipment needing to be constantly charged on a 24-hour basis (P13). Other examples of additional household costs include having supplementary bins to dispose of all of the additional waste (P4). Nappies and special needs equipment were are high cost items, nappies, especially, as the children in this study don’t move out of the nappy stage. Having to work additional hours to pay for additional household expenses was a common theme throughout, as (P4) parent describes. Others noted similar costly problems:

“When you take shopping, bills, nappies out of that, it’s hard. I manage my money well. Sensory lights cost a lot of money; anything special needs has a special cost. The extra 100.00 I get for my carer allowance goes towards those toys” [P17]

“I had to put in the meter box because the bills were large. Of course it’s hard. There’s extra’s across the board, travelling is a major thing” [P7]

(Child’s name) is on oxygen 24/7. She’s on a nebulizer. Her suction machine has to be charged. Our electricity bills are over 300.00 a month” [P13]

“We use four bins a week, we have so much waste to dispose of every week. We are paying double household charges on waste and electricity” [P4]

“Well my husband works extra hours even just to pay the bills. Travel is awful expensive. I mean last week we had to pay a couple of hundred euro on diesel to go to our appointments in Dublin and sure once you’re there you have to eat out” [P16]

Parent’s regularly reported having to adapt their car and household to adjust to their child’s needs. The additional costs incurred were often paid for privately out of family household income. Costs can be as high as €2,000, as reported by (P21) below, while another parent (P12) needed additional equipment to help support their child at home, costing up to €800:

“We had to get a loan to adapt our car. We bought a car seat that was over 2,000 euro. The thing is when buying a new car you have to have the car adapted and you need a specialized car seat. That was a major expense for us” [P21]

“The amount of stuff is endless, you are always battling with yourself whether you should get something or not. Things like shoes, it’s not like you can go into Dunnes and buy shoes for them. I have to get good supportive ankle boots. It’s hard, we’ve had to get these parallel bars, they are 700 – 800 euro” [P12]

The Medical Card

Children born with complex disabilities require many additional medications to support and manage their conditions, examples of which include: bandages, special food, as some children required tube feeding and specialized equipment, for example, nebulizers and oxygen machines (which is discussed in more detail in chapter 6). These items can add a considerable expense to families without the support of a medical card. A medical card is supplied to parents if they are under a certain income threshold, to help support with doctor’s fees and drug expenses. Parents that are granted a medical card on behalf of their child face a co-payment of €2.50 per prescription item. Applying for and obtaining a medical card is a lengthy process for parents and can ease the cost burden, as described by one parent below (P24):

“Initially we had a big battle to get the medical card. We only got only last year. We were paying 120 a month for a good while. It was hard to be doing that. We had to fill out two big forms and send three begging letters” [P24]

Even when parents do have the medical card, they can still face a significant cost due to item not being covered on the medical card, as one parent reports below (P20):

“A medical card but even with that, we’re paying up to 100.00 a month. There are things you won’t get on the medical card, like, bandages, suppositories and calpol. There’s always something not covered. A tube of cream that (child’s name) needs for his bum is

10.00. Calpol is nearly 8.00 a bottle. When he's sick, we'd use two of them a week" [P20]

The findings on the economic challenges from the qualitative study provided rationale to conduct a descriptive study on the out of pocket costs incurred by families presented in Chapter 6.

4.6.7 Master Theme 7 - Emotional Effects of Caring

Caring for a child with complex care needs is stressful and can carry many additional challenges in comparison to caring for a child that is healthy. All of the services up to this point are for the betterment of the child's needs. A service to support parent's emotional needs would be beneficial, as highlighted from the three key themes arising from the data:

- 1) Everyday Challenges
- 2) The Need for Emotional Support Services
- 3) Fear of Opening up the Wound

Everyday Challenges

A parent comments below that not only is battling for services stressful but travelling to avail of services is also a difficult challenge (P16). Children with complex disabilities, that are technology dependent, require a lot of preparation and effort to travel to appointments and this may suggest that receiving services in the home is more often a better solution for extreme cases. Sleep deprivation (P22) and feelings of powerlessness (P17) in relation to managing their child's symptoms all present challenges to parents:

"Some weeks you do feel down. You've bad weeks and good weeks. Sometimes I wonder am I little bit depressed but I would say it to the doctor" [P16]

“Some of the stress can be heat of the moment things, you might get a call that the nurse isn’t coming and you are sleep deprived and it’s tough because you are exhausted” [P22]

“Seeing a child in constant pain and you can’t do anything to help is hard but then you have that multiplied by no sleep, and your eating pattern is all over the place” [P17]

The Need for Emotional Support Services

Some parents felt this was a really valuable experience and crucial at the time when they received it. A liaison nurse from the voluntary organisation referred parents on to a counselling service when they felt that parents needed it. The role of the liaison nurse is important in such cases, as they can provide important information to parents on counselling services and also organize the support for parents to help them cope:

“The counselling was good. I did a course a few months ago, three evenings a month. It was brilliant. My liaison nurse spotted how badly I was coping and she organized the service, oh you need counselling to keep your head straight” [P24]

“I still have my bad days, the psychologist might stop you hitting a brick wall. I mean you can go four weeks here and all is grand and then you hit a wall and you’re no good to anyone then” [P8]

“I think it would be a good thing to do as a couple. It was can be a very emotional road. All the information you have in your head...it can make you feel very down. You can’t be going around crying when you have two other kids. It’s well needed” [P6]

“When I did go, it was just after he was born. I was going through the...you know...why us etc. I needed to hear it wasn’t my fault”
[P12]

The parents in this study were, in most cases, referred to a counselling service through a social worker at the hospital or a JJF liaison nurse or doctor. The need for emotional support was evident in parents who were caring for children with LLC’s and, as a result, further questions were added in phase two of this study to gather parents’ views on counselling services. The results of which are reported in chapter 6.

Fear of Opening up the Wound

Other parents had mixed emotions about attending psychological support services and expressed reservations. These include, the fear of making things worse, the fear of coming home upset, and thinking others need it more than them. In general, parents felt it wasn’t the right time for them to attend counselling as emotions were still very raw. One parent (P16) said it would be weeks before they would feel better, which reflects the level of emotional turmoil experienced.

“I think it’s just a lot easier no to think about it” [P14]

“I think if you’re going to counselling, it might mean coming home upset and I’m just about getting through the day as it is without going out to talk about it. I can’t commit to it, it would be weeks before I’d start to feel better” [P16]

“I think it might make me worse. Further down the line, maybe”
[P7]

“Ya, I think they should push you to take that help more. I suppose I’m a talker, I vent a good bit so maybe that helps. I talk to everyone

that comes in here. I think my husband needs it more than me. We can vent (women) but men keep it to themselves” [P18]

When asked what the reason was for not wanting to attend psychological support service one parent (P13) answered below:

“It’s expense and time, also I think there’s a bit of stigma attached to it as well isn’t there? Ya, you know it’s not something you’d go telling people about. They’d think, what the hell is wrong with you? (Laughs) When you’re on the other side of the fence, then it’s kind of like, well you need to talk to somebody, who’s not linked to this situation” [P13]

It is interesting that one of the reasons for not attending included the potential stigma from others. It is clear that parent (P13) cares about the views of others, nonetheless, they provide a rationale for attending the service, that is; talking to someone who is not directly involved in the situation has potential benefits.

4.6.8 Master Theme 8 - Future Concerns

The voluntary organization can only provide care to children up until the age of four years for two reasons. Firstly, in Ireland, all children at the age of four years of age are entitled to a free pre-school year and it is expected that children with disabilities will go on to attend pre-school at this age and potentially avail of other services. Secondly, the foundation cannot afford to continue funding nursing care after the age of four, as they rely on voluntary donations to fund nursing care, so a cut-off point of four years of age was agreed. In most cases however, children with complex conditions are unable to enter into the pre-school year because of the severity of their illness and require full time care at home. Parents are aware of this and have expressed concern over the continuity of care and the uncertainty of future provision under the theme:

1) A Tentative Future of Care Provision

A Tentative Future of Care Provision

Two parents (P17 and P22) have engaged, and continue to engage, in a battle to ensure continuity of care for their child. They provide financial rationale for the health service executive to continue funding care for their child after the age of four years. They feel they are saving the state money and that in the grand scheme of things, the funding they require is insignificant. Parents in this study universally revealed concerns about the future; one parent (P22) talks about how they are afraid to think about the tentative future of service provision because the thought of having no support is too much to bear:

“I know I will keep fighting for it. I will conduct a road block if it happens, I would leave (child’s name) in hospital and cost the state thousands of euro’s a week instead of the 16.00 euro an hour for my JJF nurse. All of these people on the HSE Boards have no experience with disabilities. They don’t have a feeling of how life actually works” [P17]

“I do worry if (child’s name) makes it to 4 years though. We are having a battle with them. We assumed when the JJF were unable to provide care after four years of age, that the HSE would continue to pay for the care. I can’t see the HSE continuing to pay for the care. We’re going to get a massive cut in care by losing the JJF hours. From what I’ve seen, they don’t replace those hours. You know from reading the paper. It’s with the ombudsman now. It’s only 256.00 euro a month” [P21]

“I don’t really think to the future too much. You know the four year limit is there, two years have gone by so quickly. It’s a frightening thought to think about when he turns 4. He won’t be going to

school...he'll be a home baby. It's going to be tough not having the support." [P22]

Parents in this study experience considerable anxiety over the uncertainty of care provisions for their child after four years of age, which further adds to the daily stress of caring for their child. The battle to obtain services is challenging because there are no clear pathways to procure essential services. Care plans for children after the age of four with life-limiting conditions is a grey area that needs more attention.

4.7 Bracketing in IPA

Bracketing is described by Gearing (2004) as a scientific process in which a researcher suspends or holds in abeyance his or her presuppositions, bias, assumptions, theories, or previous experiences to see and describe the phenomenon. There are many debates on bracketing in the literature, for instance Larkin et al. (2006 p. 106) states that "it is not actually possible, even if it might be desirable to remove ourselves, our thoughts and our meaning systems from the world, in order to find out how things 'really are' in some definitive sense". For the purpose of this study, an external researcher reviewed a draft of the interview protocol to ensure there was no pre-conceived assumptions embedded in the questionnaire, as recommended by (Larkin et al., 2006). This was a useful exercise, however it is my belief that the complete bracketing out of assumptions is impossible. Reflexive journaling was utilized throughout the research process to identify any preconceptions throughout the research process and is discussed in more detail below.

4.8 Reflexivity

Reflexivity in qualitative research is different to objectivity and so it is important to acknowledge that researchers are part of the world they are studying, and that their values and interests may impinge on the research work (Fade, 2004). Reflexivity requires the researcher to be perceptive to the ways in which they have shaped the

gathering and analysis of the data (Rolls and Relf, 2006) Throughout this research project, I have reflected upon and inspected my role in the shaping of the data collection and analysis process. This is essential to ensure that potential biases from the researcher's own subjective experiences are reduced. This research project is underpinned by my own personal values and shaped by my academic background in health economics and health services research. This, I believe, has led to a research project that is supportive of both service users and service planners at their respective micro and macro levels.

I personally found the initial interviews challenging to conduct, as I was coming from a non-clinical background, with no previous experience with children who had life-limiting conditions. There was a hesitance on my part to fully relax in the setting, however, as the interviews progressed, my technique improved and I felt more comfortable interviewing families. This was partly because of the warm response I received from families and their need to express their experiences. I didn't feel that parents were presenting a dramatized version or holding back on anything important. I felt they were exceedingly open and honest at all times. Extremely emotive topics were discussed during the interview process and it was difficult to forget about some of the things that were disclosed. The process of reflexive journaling helped to debrief after particularly difficult interviews.

The interpretation phase was complex but the approach to analysis proposed by Mays and Pope (2000) provided an excellent guide. During the analysis, I had to listen to the transcripts several times to ensure I fully comprehended what was being said. I hoped that the research would reveal financial challenges experienced by parents because of the base-line study I was involved in. As a result, I may have been attuned to finding this, which unavoidably leads to researcher bias, and reflects my own view on what I anticipated finding. However, several validity exercises ensued; presentation of results to the service providers, transcripts checked by a leader in the field and an independent researcher agreed on the themes, all of which mitigate for potential bias in this study. I believe this process has improved the quality of the research and that my own interpretations reflected what was conveyed and said by the participants.

4.9 Summary Discussion

Chapter four utilized a phenomenological approach to elucidate parents' lived experiences of caring for children with life-limiting neurodevelopmental conditions, with a particular focus on their service use experiences. The eight key emergent themes were:

- 1) Transition from hospital to home
- 2) The perpetual wait for services
- 3) The crucial role of the liaison nurse
- 4) Parents as advocates
- 5) The conflict in obtaining services
- 6) Physiotherapy - a very valued service
- 7) The economic costs of caring
- 8) Future concerns

Parents' experiences of using services can be characterized by long waiting times, on-going battles with service providers, and uncertainty about the continuity of care. Experiences of hospital-based services were inconsistent with issues of poor communication and discharge management. Community care services were strictly allocated and parents faced extremely long waiting times for service-set up. The disconnect between statutory and community care services was prevalent, while the significant role the voluntary organization plays in filling service gaps in the community is evident. While most all parents found the quality of the service to be satisfactory, the battle in obtaining services could be described as a constant everyday struggle. What was also evident was the significant and worthy role of the link worker, which when provided to families, had insurmountable benefits for the well-being of the child and family.

In this chapter, the findings of phase one of the study are reported, which examined parent's lived experiences of caring for children at home with life-limiting neurodevelopmental conditions, with a particular focus on their service use experiences. A detailed discussion on these findings is presented in Chapter 7.

4.10 Strengths and Limitations of the Study

Traditionally, IPA studies are conducted with small samples sizes, so the relatively large sample used in this study may result in missed nuances in the data. It was not possible to discuss all of the emerging themes but the ones that were discussed were representative of the population studied. Some challenges must be acknowledged, including the need to recognise that interpretations are challenged by a participant's ability to adequately articulate their thoughts and experiences Smith et al. (2009) and that the interpretative nature of the research was influenced by the researchers pre-existing knowledge and perspectives. This was addressed however, through ensuring the findings were grounded in data and a range of inter-rata agreement exercises were conducted. The findings in this study are congruent with other research on parents caring for children with complex care needs in the community (Baillie et al., 2000). Most of all the findings from the IPA analysis highlights the need to conduct more research to bridge the gap between parent's expectations and those of service providers.

Chapter 5

Preferences of care services for children with life-limiting neurodevelopmental disabilities in Ireland - a discrete choice experiment

5.1 Introduction

Making decisions on the appropriate choice of alternative models of service provision requires an assessment of their associated costs and benefits, of which there is a lack of evidence internationally (Emond and Eaton, 2004). Governments and voluntary organisations are increasingly interested in public and patient preferences, for both informing policy agendas and for aiding decision-making. Previous research in this area (mostly UK based) has typically focused on identifying parents' care needs, using qualitative-based survey methods. Scarcity, coupled with the need to make choices between competing claims on resources, has focused attention on economic evaluation (Lancsar, 2008). This study applies a quantitative methodology, namely a discrete choice experiment (DCE) to elicit parents' preferences for care surrounding services for their children who have life-limiting neurodevelopmental conditions; to the best of our knowledge this quantitative methodology has never being applied to this particular population group. Applying a quantitative methodology to this group will provide a unique insight into the strength of preferences for particular service types, and for the relative importance of various service types and their marginal rates of substitution. The findings will provide a new evidence base to make specific recommendations to policy makers and inform service planners on how best to meet parents' needs.

The purpose of this chapter is to introduce and provide motivation for phase two of the study. A brief overview of Discrete Choice Methodology (DCE), and its theoretical underpinnings, is outlined and the stages in the development and design of the DCE procedure is delineated. The results section has subsections in which different models are ascribed to the data (i.e. differing assumptions are made).

These different models agree broadly on the main findings. The chapter concludes with a summary of the findings and limitations of the study.

5.2 Phase Two Objectives

5.2.1 Study Aims:

- Quantify parents' relative preferences for health care services for their child at home.
- Quantify trade-offs i.e. how much of one service would parents be willing to trade for another.
- Estimate monetarily how much they value particular services.
- Determine which socioeconomic and demographic characteristics are related to parental preferences for care services in the community.

5.2.2 Potential Study Outcomes:

- Support decision-making in the allocation of services offered to children with complex disabilities living at home.
- Establish what parents need more of, in relation to proving care for their child.
- Identify a monthly package of care that is suitable for children with LLC's.
- Provide original quantitative estimates for this population group.

5.3 Discrete Choice Experiment Methodology

5.3.1 An Overview of Choice Methods used by Economists

Discrete choice experiments are an attribute-based measure of benefit, based on the assumption that health-care interventions, services or policies can be described by their attributes (Ryan and Gerard, 2003). They involve the generation and analysis

of choice data, and the creation of hypothetical scenarios that can be constructed to suit relevant research questions (Lancsar, 2008). Economists differentiate between two approaches of measurement: 1) revealed preferences (observed behaviour) examples include, the travel cost method and the hedonic pricing method and 2) stated preferences (derived from surveys & questionnaires) e.g. contingent valuation methods (CVM) and Discrete choice experiments (DCE's) (Bridges et al., 2011). These are discussed in more detail in the following two sections.

5.3.2 Revealed Preference Technique

Revealed preference (RP) techniques presents participants with information on current market equilibrium, which can be used to forecast short-term departures from existing circumstances; RP yields one observation per participant and technological constraints are fixed (Louviere et al., 2000). There are criticisms of the RP technique however; one such example, is that it is impossible to infer consumer values or preferences from RP data (Ryan et al., 2007). Also, given that many aspects of health care are not traded in markets, i.e. there is a zero cost at the point of service and they have public-good characteristics, it is difficult to elicit unbiased estimates (Ryan et al., 2007).

5.3.3 Contingent Valuation Method

The Contingent Valuation Method (CVM) asks individuals directly how much they would be willing to pay (WTP) for a particular good or service. It is traditionally used to value goods with public good characteristics that have a zero cost associated with it (Mitchell and Carson, 2013). Examples include the willingness to pay for water, the prevention of an oil spill, or a public park. It essentially asks people to provide a monetary value for something they do not already pay for. It is not without its criticism, due to its lack of scope of sensitivity, warm glow effects, and that it only elicits cost based responses (Ryan et al., 2001). Moreover, depending on how the CVM questions are framed, i.e. direct questions or bidding, issues of starting point bias or range bias can happen, leading to less robust WTP estimates (Klose, 1999).

5.3.4 Discrete Choice Experiment Method

In contrast the methods outlined above, DCE's allows one to design realistic scenarios *a priori* and control the stimuli that generate the data (Reed Johnson et al., 2013). DCE's also have the ability to predict behaviour changes and trade-offs over a much wider range of attribute levels than the current market provides. An added strength of DCE's, is that they extract numerous responses per participant and essentially force participants to make trade-offs between the various scenarios under consideration. The results provide useful information on participants' relative preferences for services, and on their willingness to trade between services. The results of DCE's reveal how marginal changes to the delivery of services will impact health care utilisation. The estimates from the DCE can also be used to estimate preferences in the context of cost benefit analysis and, subsequently, policy analysis. Some criticisms of DCE's include the cognitive burden to participants of having to "trade" between many attributes and levels and this was a particular concern for the chosen population of this present study. Reducing the number of choice tasks presented is one way to counteract this issue but this then limits the amount of information you can generate from the experiment at any one time (Bryan and Dolan, 2004). A critical phase of DCE's is the design stage where measures can be taken to overcome these challenges; this is further discussed later throughout the design stage.

5.3.5 A Brief Overview of Discrete Choice Experiments

DCE's are typically implemented in surveys comprising several choice sets, each containing hypothetical options from which respondents choose. Each option is described by a set of attributes and each attribute takes one of several levels. Levels describe ranges over which attributes vary. For example, in this study, we elicit parents' preferences for care services for children with LLC's. Parents were presented with a range of choice scenarios, each containing varying amounts of particular services. They consider the varying attributes and levels and then chose the package that yields the highest utility. In DCE's, respondents make decisions

about quantity or quality differentiated versions of a good or service that requires them to make trade-offs. The resulting choices are analysed to estimate the overall utility and willingness to trade between services. DCE's have been increasingly applied to health care in the last decade, which is likely to do with the fact that they can elicit patient's values on all aspects of care. The QALY (Quality Adjusted Life Year) in comparison does not consider patient preferences, just QALY weights, hence the increasing pressure on the National Institute for Clinical Excellence (NICE) to take account of patients' preferences (Ryan, 2004, Birch and Gafni, 2002).

5.3.6 Overview of DCE in Health Services Research

A recent systematic review on the application of DCE's to health and health care highlighted key areas of application which includes: market choices amongst health care professionals, priority setting frameworks at the local/national level, and preferences regarding clinical decision making (de Bekker-Grob et al., 2012). Examples of topic areas include preferences for drug treatments for: juvenile arthritis (Burnett et al., 2012), osteoporosis (de Bekker-Grob et al., 2008, de Bekker-Grob et al., 2009) and osteoarthritis (Fraenkel et al., 2014). Also investigated were: physician service preferences in the Ukraine (Danyliv et al., 2014), preferences relating to continuity of care in GP practice in the UK (Turner et al., 2007) and physicians' preferences for prescribing in different care contexts in Denmark (Pedersen et al., 2014). Preferences for cancer screening is another key area of application and in particular, the area of colorectal cancer screening (Benning et al., 2014, Hol et al., 2010, Howard and Salkeld, 2009, Marshall et al., 2007, Hawley et al., 2008).

5.3.7 Application of DCE's to Palliative Care

Studies on preferences for end of life care using DCE's are limited and existing studies are predominantly focused on the adult population. Some examples include: preferences for end of life care services for adults in four palliative care day centres in England (Douglas et al., 2005), concordance of preferences for end of life care

services for adults with advanced stage cancer and their caregivers in Singapore (Malhotra, 2014) and finally, patient and professionals preferences for organ allocation and procurement for patients with chronic kidney disease at end of life in the USA (Davison et al., 2010).

5.3.8 Application of DCE's to Paediatric Health Care

Only three studies appear in the literature eliciting parents' preferences for care services for their children. These include: dual preferences of parents and anaesthesiologists on paediatric daycase surgery (Gidman et al., 2007), parents' preferences for care services for children with mental health problems (Urey and Viar, 1990) and parents' preference for epilepsy diagnostics (Wijnen et al., 2014). This study is the first of its kind to apply a DCE methodology to children with life-limiting neurodevelopmental disabilities. In doing so, it provides a unique contribution to the literature on paediatric palliative care services.

5.4 Theoretical Underpinnings of Discrete Choice Experiments

DCEs are grounded in Lancaster's economic theory of value (Lancaster, 1971), where it is assumed that the demand for a particular good or service depends on the characteristics (attributes) of the good or service, rather than the goods themselves. Within a choice experiment, respondents are asked to choose between two or more alternatives; the resulting choices reveal an underlying (latent) utility function (de Bekker-Grob et al., 2012). In each choice scenario, it is expected that individuals will contemplate all information offered and then choose the alternative that yields the highest utility (Ryan et al., 2007). Choice data is then analysed using McFadden (1973) the random utility maximisation framework (RUM).

5.4.1 Random Utility Model

The basic random utility model, for party 1 ... n ... N and choice $i = A$ or $i = B$, states that the utility for party n picking package i is given by

$$U_{ni} = V_{ni} + \epsilon_{ni}$$

where V_{ni} is a deterministic function of the package attributes and ϵ_{ni} is a random function of unobserved service attributes and individual variation in preference. The systematic, explainable portion of utility, denoted V_{ni} , is modeled as

$$V_{ni} = \beta_1 x_{1i} + \beta_2 x_{2i} + \dots + \beta_m x_{mi}$$

where x_{mi} is the level of attribute m in package i . Note how V_{ni} is assumed to have a linear form. This does not appear to be, too restrictive an assumption (Ryan and Gerard, 2008), although it is not uncommon to have a quadratic utility component associated with a salary variable (Kolstad, 2011). In our study, participants chose between two unlabelled (i.e. generic) choice sets, A or B. For this reason, it was not an expectation to see alternative-specific effects, hence the omission of a constant term α_i in the specification of V_{ni} above.

Party n will choose whichever package maximises their utility; in other words, the probability of picking a package is given by its relative utility versus any alternative:

$$P_{nA} = \Pr(U_{nA} > U_{nB})$$

$$P_{nA} = \Pr(V_{nA} - V_{nB} > \epsilon_{nB} - \epsilon_{nA})$$

The left hand side of the inequality above is known and deterministic - what remains to determine P_{nA} is the difference of the unobservable error terms. At this point, assumptions about the distribution of error terms must be made. Since probabilities must add up to one, there are various constraints on types of error distribution. In this present study, all variables x are treated as continuous variables.

5.5 Study Participants and Sampling

As already discussed in Chapter 3 the participants in this study were parents of children under the age of seven years of age with life-limiting neurodevelopmental disabilities. The final sample of agreed participants (Chapter 3 Sampling Frame)

was 104 families. Determining the sample size for the choice experiment is discussed in the following two sections.

5.5.1 Sample Size

According to Lancsar & Louviere (2006), sample size in a DCE is contingent on the following factors: the true values of the unidentified parameters, experimental design i.e. the number of attributes and levels in the design, and the resultant number of choice sets. Clear guidelines are lacking in the literature on methods to calculate sample sizes for DCE's.

5.5.2 Sample Size Formula

We adopted the formula used by Orme (1998) to estimate the sample size necessary to achieve a tolerable margin of error (M.O.E.), using a 95% confidence interval (CI)

$$\text{M.O.E.} = \pm 1.96 \sqrt{\left(\frac{pop-ss}{pop-1}\right) \frac{pq}{ss-1}}.$$

Where *pop* refers to population size, *ss* refers to sample size and *p* and *q* are the expected probabilities of choosing package A and package B respectively (typically a worst-case assumption of $p=q=0.5$ is made). This analysis suggests that, for small populations, a high proportion is needed to make up the sample. Inserting 280 for the population and 69 (i.e. an approximate response rate of 50-60% of total population) for the sample size gives a 10% margin of error for our statistical estimates. While this is not ideal, this is to be expected using the above worst-case analysis. Empirically, previous studies have found that a sample size of 30 and, later, 75-100, was adequate. Also, for investigational works and hypothesis generating studies, a sample size of between 30-60 respondents is sufficient (Orme, 1998). Given the circumstances in which parents find themselves, the proportion of

the population that completed questionnaires was in fact relatively high.

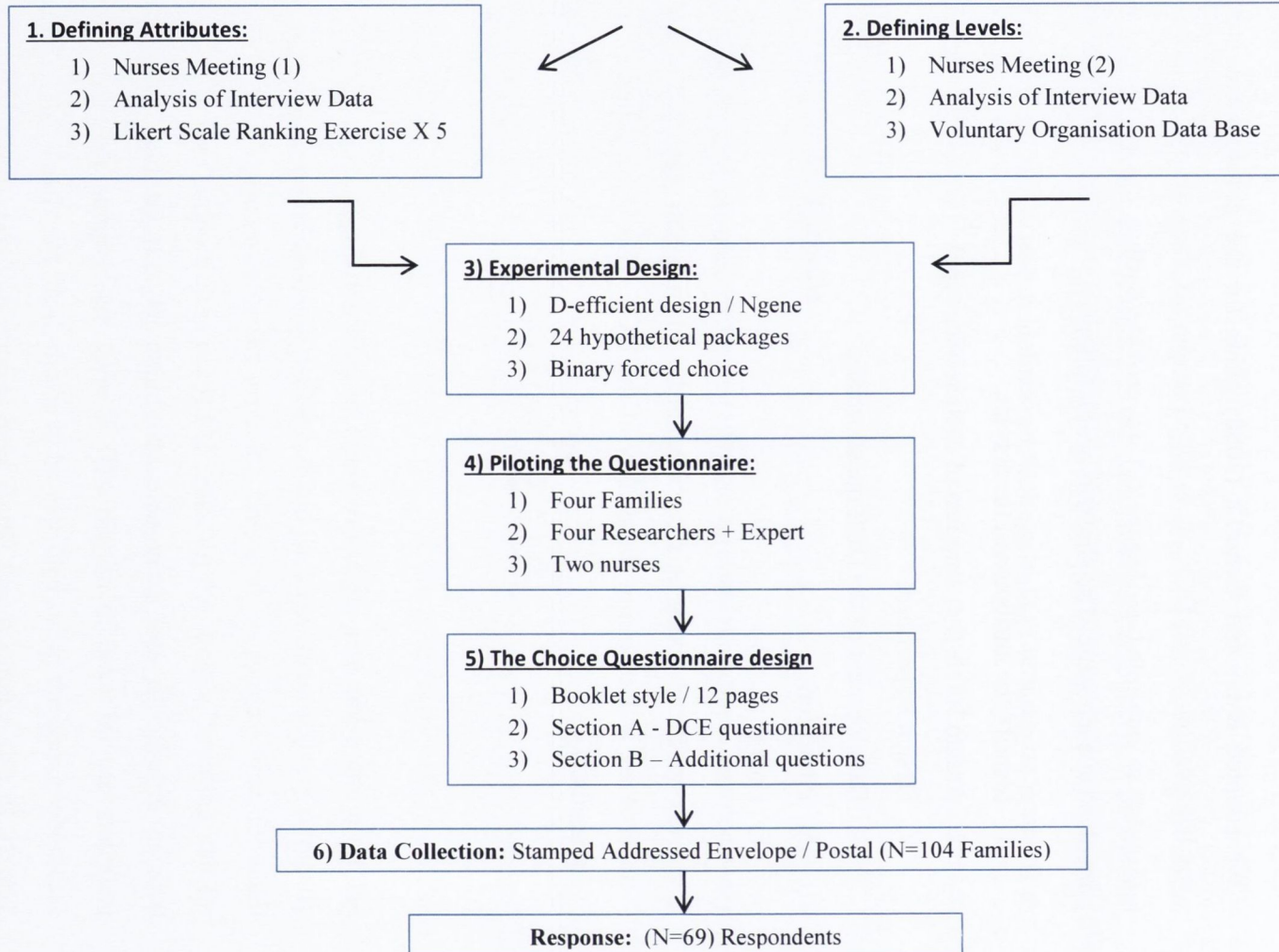
5.6 Design of Discrete Choice Experiment

We adopted Ryan and Gerard's (2008) guide for the development, design and implementation of the DCE, as outlined in the six steps below. A more detailed diagram is presented in Figure 5.1 to provide further clarity on the activities involved at each stage. The six steps are as follows:

1. Identify the attributes of the service
2. Assign levels (i.e. amounts of each service type)
3. Design Experiment
4. Development of the DCE questionnaire
5. Pilot Work
6. Data collection

In addition, the checklist by (Lancsar and Louviere, 2008) was used as a guide for undertaking and assessing the quality of the DCE. This checklist is reported in Appendix (14).

Figure 5.1 Stages in DCE Design



5.7 Identifying Attributes

The identification of a set of attributes should be determined by factors that will affect respondents' choices, along with those attributes that are policy relevant (Ryan and Gerard, 2008). According to the conjoint analysis checklist for good practice from the International Society for Pharmacoeconomics and Outcomes Research (ISPOR), three criteria should be considered when deciding which attributes to include in the DCE: 1.) applicability to the research question, 2.) significance to the decision context, and 3.) whether attributes are correlated to each other (Bridges et al., 2011). Qualitative work has been more commonly applied to identify attributes and enhance the face validity of DCE's. Methods used to develop attributes and levels typically include: literature reviews, focus groups, interviews and expert opinion. Coast (2012) considers the gathering of opinions, from those that have direct experience with the context under investigation, a very valuable exercise. Attributes informed by qualitative research are often richer and, in addition, are useful for discussing sensitive subjects, which was particularly relevant for this study.

5.7.1 Nurses Meeting (1)

The first step in defining attributes involved an open discussion with eleven nurses from the JJF voluntary organization, to obtain a list of all services typically utilised by parents and to obtain their child's frequency of service utilisation on a monthly basis. A detailed list emerged, containing 20 different service types, as outlined in Table 5.1. DCE's seldom incorporate all of the important attributes, but it is necessary to include the most essential ones pertinent to the majority of respondents (Kløjgaard et al., 2012). A number of tasks were conducted to refine the large service list and this is presented in the following three sections.

5.7.2 Contribution from Phase 1 Interview Data

Data from the 24 interviews conducted in phase 1 was used to confirm and refine the service list obtained at the nurses meeting. Information was gathered from parents on the types of services they most commonly utilised, the frequency of service use and the quantity of each service they would typically receive e.g. 1-2 hours of physiotherapy in a given month. Deductive thematic analysis was used to further refine the list of service types. Services that were not utilised on a regular basis, and which were believed to be unimportant by parents, were removed from the list. The final list consisted of eight different types of services (see Table 5.1). Presenting parents with eight different service types to choose from could potentially cause respondent fatigue, poor response efficiency, and later, measurement error (Reed Johnson et al., 2013). In anticipation of this, a Likert scale ranking exercise was presented to reduce the list of attributes.

5.7.3 Likert Scale Ranking Exercise

To facilitate the construction of the final list of attributes, a ranking exercise to assess the importance of attributes, as recommended by Bridges et al. (2011). A convenience sample of ten families (i.e. 10% of agreed study participants n=104), were approached and asked to participate in the exercise. It is important to note, that before contacting participants throughout all stages of this study, contact was made with the foundation to verify that it was an appropriate time to contact families. Children with such conditions regularly fall ill and therefore it was important to avoid contact families at a sensitive time. All families expressed interest in participating but only five actually completed the ranking exercise. This exercise was completed face to face alongside the researcher in family homes. All of the families that participated in the exercise had participated in the interviews and had familiarly with the researcher as a result. A Likert scale (Appendix 15) was presented to parents to investigate heterogeneity between service preferences (Breffle et al., 2011) and is a useful way to obtain more detailed information and to sharpen the choice model representation (McFadden, 1986).

The five-point scale ranged from least preferred to most preferred, which

included eight different service types. One of the eight service types included a counselling service for parents, as during the interview process, parents expressed a need for this service. We also added a hypothetical cash maintenance payment. The cash payment was added for three reasons: 1) it serves as a proxy for other items that can help with some unmet needs, 2) it can serve as compensation for the cost burden facing families, and 3) it can estimate the willingness to pay (or willingness to accept compensation for a loss of one particular service). The cash payment was defined as a monthly payment that could go towards household expenses. Evidence from a UK survey found that parents caring for disabled children prefer to receive regular cash payments instead of a once off annual payment (Beresford, 1995, Redmond and Richardson, 2003).

5.7.3.1 Results from Likert Scale Exercise

All families had a clear preference for six of the services presented; preferences did however vary for out of home respite services but it was decided that this was still an important attribute, given all the data that was collected. Parents had a high preference for a counselling service but as this is essentially a service for parents and not children, it was decided that a separate question would be added to assess parents' value for it (the results of which is presented in Chapter 6). Fieldwork conducted during phase one revealed differences in preferences for night and day nursing care, with some families preferring to have night nursing, while others preferred day nursing care. It was decided to treat both night and day nursing as two separate attributes, as combining both attributes to one overall nursing care service could result in a loss of information (Kløjgaard et al., 2012).

Six attributes were assumed to be a suitable number to present to participants, as anything above that would become too cognitively demanding and particularly so for this population (Ryan and Gerard, 2003). Not all attributes can be included in a choice experiment; the decision to use six and not eight was ameliorated by adding additional questions on the services that were excluded. In section two of the choice questionnaire, further questions were asked on parents views of counselling services and their willingness to pay for this service. Furthermore, counselling services are a direct support to parents themselves, so it

was decided to specifically focus on the services that were directly related to the needs of the child. The results of the additional questions are presented in Chapter 6. The final six attributes to use in the experiment were: night nursing care, day nursing care, physiotherapy, speech & language therapy, out of home respite care, and a cash maintenance payment, outlined in Table 5.1

Table 5.1 Refinement of Attributes for Study

Original Service List (N=20)	Reduced List (N=11)	Final List (N=6)
Nursing care	Night Nursing	Day Nurse
Carer	Day Nursing	Night Nurse
Physiotherapy	Physiotherapy	Physiotherapy
Occupational Therapy	Occupational Therapy	Speech & Language
Speech & Language	Speech & Language	Cash Payment
Social Worker	Respite out of home	Respite out of home
Disability Liaison	Cash Payment	
Public Health Nurse	Info from Liaison Nurse	
GP	Info from Social Worker	
Counselling Services	Counselling Service	
Play Therapist		
Home Teacher		
Dentist		
Orthotics		
Special Needs Assistant		
Paediatric Outreach Nurse		
Emergency Services		
Public Health Nurse		
Community Welfare Officer		

Respite out of home

5.8 Attribute Levels

A good experiment is one that has a suitably rich set of attributes and choice situations, together with enough difference in the attribute levels to generate important behavioural responses (Ryan and Gerard, 2008). Levels in particular should reflect the range of situations respondents might expect to experience (Mangham et al., 2009). Levels were defined as the amount of a particular service a family receives; the unit of measurement for this was hours, for example, physiotherapy ranged from one visit (one hour) to 3 visits (three hours) in a given month and so on.

5.8.1 Contribution from Phase 1 Interview Data

Fieldwork informed the levels used for the experiment, which consisted of base levels (i.e. current practice) and all additional levels represented improvements from the base, which were broad but realistic. Providing realistic and feasible levels increases the precision of the parameter estimates. The cash payment reflected realistic payments that the voluntary organisation could potentially provide; it was also comparable to the annual statutory grant that parents receive.

5.8.2 Voluntary Organization Data Base

Given that the JJF provide funding to families to purchase nursing care, levels were checked against real data provided by the voluntary organization database.

5.8.3 The Number of Levels

Three levels for each service type were decided upon, which was in keeping with current circumstances facing families. The larger the number of levels assigned to an attribute, the more likely significant values will be obtained, as respondents tend to place more importance on attributes with extra levels (Ratcliffe and Longworth,

2002). Also, having any more than three levels would increase the number of possible scenarios required exponentially (Ryan and Gerard, 2008). A critical assumption in DCE's is "unlimited substitutability" between attributes i.e. when choosing one service type over another, it is important that participants are compensated for the loss in one level while staying with the same level of utility in another (Ryan and Bate, 2001). Moreover, unlimited substitutability ensures more even indifference curves for the calculation of marginal rates of substitution (Deaton, 1980, Ryan and Bate, 2001).

5.8.4 Nurses Meeting (2)

A second and final meeting was held with the JJF nurses to confirm all attributes and levels were in line with current practice. Strong communication with the voluntary organisation was important during this phase to ensure realism and plausibility in the experiment, as recommended by Ryan and Gerard (2008). This was a useful exercise, as the amount of nursing hours allocated to families had recently been reduced from 80 hours to 60 hours monthly. This was a result of reductions in voluntary contributions made to the organization during 2012. The required changes were made to reflect this reduction in nursing care. Monthly nursing hours were reduced from 100 hours to 80 to reflect this change. The final list of attributes and levels are reported in Table 5.2.

Table 5.2 Table of Attributes and Levels

Attributes	Description	Levels (coding used in analysis)
Night Nursing	Monthly hours of night nursing	18 hours monthly (18) 24 hours monthly (24) 32 hours monthly (32)
Day nursing	Monthly hours of day nursing	30 hours monthly (30) 55 hours monthly (55) 80 hours monthly (80)
Physiotherapy	Monthly hours of	1 hour monthly (1)

Attributes	Description	Levels (coding used in analysis)
	Physiotherapy	2 hours monthly (2) 3 hours monthly (3)
Speech & Language	Monthly hours of S & L	1 hour monthly (1) 2 hours monthly (2) 3 hours monthly (3)
Respite out of home	Monthly days in out of home respite facility	1 day (12) 2 days (24) 3 days (36)
Respite out of home	Monthly days in out of home respite facility	1 day (12) 2 days (24) 3 days (36)

5.9 Experimental Design

The objective of an experimental design is to extract the maximum amount of information from each choice situation. The combination of attributes and levels (six attributes with three levels) yielded $3^6 = 729$ possible combinations i.e. a full-factorial design. A full factorial design has the benefits of allowing an estimation of all interaction effects (Ryan and Gerard, 2003), however, for practical reasons and fear of respondent fatigue, a fractional factorial design was used. At the experimental design stage, a smaller set of choices are selected which enables main effects and interaction effects (de Bekker-Grob et al., 2012). The best-known fractional factorial design is the orthogonal design, which aims to minimise the correlation between attribute levels in the choice situation. One criticism of this design is that it increases the chance of presenting participants with a dominant alternative.

5.9.1 D-Efficient Designs

More recently, statistically efficient designs are used in DCE's, which are said to outperform orthogonal designs (Street et al., 2005, Street and Burgess, 2007). Experimental designs that incorporate priors can be statistically more efficient than designs that do not. These priors could be estimated from previous studies, pre-test data, pilot test data or logic (Reed Johnson et al., 2013). Efficient designs rely on prior knowledge of the parameter estimates, which we had obtained from open discussions with experts, in-depth interviews and face-validity exercises. Prior values were incorporated into the design using *Ngene* software (Choice Metrics, 2012). Very small, but positive, parameter estimates for each of the attribute coefficients had the effect of removing dominated alternatives from our DCE design. Such dominated alternatives are when one package is superior to another in every way; these have sometimes been argued to be useful tests for rationality, but with 12 choice sets, it was important that each choice set contributed to the data that will be analysed. A common measure of statistical efficiency is the D-efficient criterion, where the determinant of the variance co-variance matrix is minimized (de Bekker-Grob et al., 2012).

5.9.2 Main Effects Design

A main effects design was created resulting in 24 hypothetical packages of care. The 24 care packages resulted in 12 individual choice sets. This was thought to be a feasible number to present to participants, given that previous research found that cognitive burden increases past a certain number of choice sets (Bech et al., 2011). Each choice set had two hypothetical unlabelled monthly care packages: package A and package B i.e. a binary forced choice combination, with each package varying from the other. In recent years, experiments are providing participants with “opt out” and “neither choice” but for the purposes of realism, it was more appropriate to use a forced choice, as each care package option was a better choice than opting out (i.e. no package of care).

5.9.2.1 Syntax for Ngene Design

The Ngene syntax used, is presented in Figure 5.2

Figure 5.2 Ngene syntax used for design

```
;alts=package1*,package2*

;rows=12

;eff=(mnl,d)

;model:
U(package1)=b2[.02]*nn[16,24,32] + b3[.02]*dn[30,55,80]+ b4[.02]*phys[1,2,3] +
b5[.01]*sl[1,2,3] + b6[.01]*cash[50,100,150] + b7[.01]*resoh[1,2,3] /
U(package2)=b2*nn + b3*dn+ b4*phys + b5*sl + b6*cash + b7*resoh $
```

5.10 Piloting the DCE Questionnaire

The main goal of this exercise was to obtain feedback on comprehension, interpretation, language, structure and flow (Kløjgaard et al., 2012). Parents raised some issues with the language used to present the choice tasks and called for more clarification and simplification of language in the instructions section. Bridges et al (2011) recommends simplifying the tasks as much as possible and that readability should be assessed from the study participant’s perspective. Think aloud exercises and de-briefing were used to enhance participants understanding of the choice tasks, as these are more commonly used in DCE’s (de Bekker-Grob et al., 2012).

5.10.1 Pilot Exercise with Families

An additional random sample of parents (n=10) were contacted and asked to participate in the pilot exercise. It was extremely challenging to organise suitable times with parents, and although all parents expressed interest in participating,

many cancellations occurred due to the nature of their child's illness. Some families agreed to receive the DCE by email but not surprisingly, none of the emailed DCE's were returned. After many follow-up phone calls, four meetings were arranged with families to participate in and review the pilot DCE.

5.10.2 Pilot Exercise with Researchers

To further enhance the quality of our questionnaire, and because of the poor pilot participant rate (3.8%), four researchers in the field of health services research were asked to complete the pilot questionnaire. Comments on the structure of the questionnaire and suggestions on lay out were recommended. In addition, the advice of an expert in the field of qualitative research was acquired to enhance the presentation of the questionnaire. The following suggestions were made: provide a colour print questionnaire, present the title of the study in large easy to read font, use a study acronym, i.e., "POC STUDY", and present the questionnaire in booklet style format with official Trinity College Dublin (TCD) and Jack and Jill Foundation (JJF) logos on the front. It was also suggested that we provide a large section for feedback. All suggestions were incorporated into the final design.

5.10.3 Nurses Feedback

Two questionnaires were sent to two expert nurses at the foundation for a final review to ascertain that all questions were sensitive and deemed suitable for families. The nurses agreed all questions were sensitively worded and appropriate. A timeframe for pilot exercises was set and all pilots were completed and analysed within approximately eight weeks (April 1st – June 1st 2013).

5.11 The Choice Questionnaire

The DCE questionnaire was titled "*Questions about parents' preferences of care services for children with complex care needs in Ireland*" (as presented in Appendix 13). The term "complex care needs" was decided as the best term to use, as it was

broad and encompassed the heterogeneous diagnoses in this study. Moreover, specifically using “neurodevelopmental diagnosis” would exclude those who have not yet received a diagnosis. The questionnaire commenced with a letter to parents describing briefly the goals and objectives of the study and a reminder that the information provided would be treated as strictly confidential. This was followed by a brief description of section one (choice questionnaire) and section two (additional demographic data) of the questionnaire. Pilot exercises took approximately 10-15 minutes to complete, so this was the length of time suggested to participants in the instructions section. Contact details (i.e. mobile number and contact address) were provided should for parents to raise any questions or concerns about the questionnaire.

Detailed instructions on how to complete the choice tasks and a description of attributes and associated levels followed. Parents were presented with 12 choice sets each containing two unlabelled hypothetical monthly care packages: package A and package B. Parents were asked to select the care package that they most preferred. Figure 5.3 presents an example of one of the choice tasks presented to parents in the DCE questionnaire:

Figure 5.3: Example of Choice Task Presented to Parents

Choice set 6	Monthly care package A	Monthly care package B
Night nursing hours	24 hours	24 hours
Day nursing hours	55 hours	55 hours
Physiotherapy hours	3 hours	1 hour
Speech & Language hours	1 hour	3 hours
Respite out of home	3 days	1 day
*Cash maintenance	€150	€50

Choice A or B? (Please tick one) A B

Following completion of the choice tasks, parents were asked to report ease of completion of the DCE questionnaire, ranging from “not at all difficult” to “extremely difficult”.

5.11.1 Section Two of DCE Questionnaire

In section two of the DCE questionnaire, parents were asked about their economic situation which included questions on weekly household income, and the financial out of pockets costs associated with caring for their child. Other questions were asked about the need for emotional support services, particularly counselling to help families cope. The results of section two are reported in Chapter 6. Also, in section two, parents were asked to provide information on socio-demographic characteristics, including, diagnosis of the child, if they are caring for more than one child with a disability, marital status, family size and geographic location. All of this data is presented in table 5.3 below.

5.12 Data Collection Method

Data collection requires consideration of the sample size, mode of administration and ethics (Bridges et al., 2011). A self-complete postal survey is the most commonly utilised method in DCE's (Lancsar and Louviere, 2008) and was the most efficient and cost effective mode of administration for this study. Before contacting any of the families, the voluntary organisation was contacted to inform the researcher of any deaths that had occurred. Sadly, nine children from the study sample had died. To ensure ethics were adhered to, all of the data belonging to decedents were removed from the database to ensure they did not receive a questionnaire or any form of contact throughout the duration of the study. The remaining sample size was $(104 - 9 = 95)$.

5.13 Time Frame

To ensure response efficiency, a letter from the organisation's chief executive was attached to the choice questionnaire, encouraging parents to participate in the study. No incentives to complete the choice questionnaire were offered to the participants. The questionnaire was sent to 95 families with a stamped addressed envelope for return. Parents were asked to return the fully completed questionnaire within one month from the date it was sent out (June 30th - August 30th 2013). Thirty questionnaires were returned by 30th of August 2013. Follow up phone calls were made to families to encourage them to return questionnaires; this was a productive

exercise, as an additional 39 questionnaires were returned by October 2013.

5.14 Responses

Of the 95 questionnaires, 69 fully completed questionnaires were returned (a 72% response rate) with all 12 choice tasks completed, thus, the number of individuals providing responses for the regression analysis was 69 yielding 828 completed choices, resulting in 1,656 observations. Out of the 828 choice sets, 410 “Package A” choices were made and 418 “Package B” choices were made, confirming the validity of our choice to omit a constant term from our unlabelled choice experiment. Furthermore, the balance of choices between the package A and B shows that families acted rationally when selecting choices in the experiment. The majority of respondents did not express difficulty in the completion of the questionnaire e.g. (n=25) found it “not difficult at all”, (n=38) “only moderately difficult”, (n=4) found it “very difficult” and finally (n=2) respondents found it “extremely difficult” to complete. The respondents who found it extremely difficult did not provide further commentary as to why they did. Complexity and the challenge for participants is an issue reported in the DCE literature (Boxall et al., 2009, Louviere, 2006) and raises the question if choices were answered correctly. Upon inspection of the data however, it appears that rational choices were made as already stated above. Table 5.3 reports the respondents and children’s characteristics.

Table 5.3 Respondent and Children’s Characteristics

Sample characteristics	(N = 69)
<i>Age:</i>	
<i>Age of children (mean)</i>	2.8 (1-7)
1 year	3 (4.3%)
2 years	28 (40.5%)
3 years	19 (27.5%)

Sample characteristics	(N = 69)
4 years	13 (18.8%)
5 years	4 (4.3%)
7 years	2 (2.8%)
<i>Marital status:</i>	
Married	51 (73.9%)
Living with partner	10 (14.4%)
Single	2 (2.8%)
Separated	5 (7.2%)
Civil Partnership	1 (1.4%)
<i>Location:</i>	
Rural	47 (68%)
Urban	22 (32%)
<i>Diagnosis</i>	
Confirmed diagnosis	60 (86.9%)
Diagnosis unknown	9 (13%)
>1 child with disability:	8 (11.5%)
<i>Primary carer:</i>	
Mother	51 (72%)
Father	4 (5.7%)

Sample characteristics	(N = 69)
Shared (parents & child-minder)	11 (14.4%)
Other i.e. child minder / grandparent	3 (4.3%)
<i>Disposable household income per week:</i>	
< 50	1 (1.4%)
50 - 100	1 (1.4%)
101 - 250	9 (13%)
251 - 500	24 (34.7%)
501 - 750	15 (21.7%)
751 – 1,000	7 (10.1%)
1,001 – 1,500	10 (14.4 %)
1,500 >	1 (1.4%)
<i>Employment:</i>	
Did not give up work	12 (17.3%)
Reduced work hours	9 (13%)
Gave up work	45 (65.2%)
N/A	3 (4.3%)
<i>Medical card status:</i>	
Medical card	61 (88%)
No medical card	8 (11.5 %)

Sample characteristics	(N = 69)

5.14.1 Respondent Characteristics

As summarized in Table 5.3, the sample is quite representative of families for key measures such as diagnosis, age, and income, with the exception of location (discussed in more detail below). The children’s age ranged from 1-7 years, with 91% of the sample under four years and only 7% between the ages of 4-7. Due to financial constraints, the foundation can only afford to provide care to children up until the age of four years, however, in observing responses, one can see that in five cases, the foundation continued to finance care.

Geographic location was defined using self-reported addresses, however, in some cases, the addresses reported were vague and some responses left blank. A unique identifier was used to trace and confirm participant’s addresses. Responses were largely from rural geographic locations (68% from rural and 32% from urban areas). The location variable is complex because it serves as a proxy for many things; income is systematically linked to location, access to services, family structure and the likelihood of other families living in the area to provide support. Also, a family living in Kildare with a rural address would have quicker access to Tallaght hospital in comparison to a family living in Dublin city centre (i.e. an urban area). Thus, the challenge with location as a variable is that it is not sufficiently discriminating enough to capture the myriad of different things it represents. We hypothesized *a priori* that there would be differences in preferences based on geographic location, as some families expressed concerns over access to services based on location during the interview process, however, results from the analysis were treated with caution.

Individual diagnoses were not presented in order to protect the participant’s identity, as some diagnoses are the only known cases in the world, thus making participants easily identifiable. The most widely reported diagnoses amongst those who had a confirmed diagnosis (n=60) were Cerebral Palsy and Classical

Lissencephaly 1-24, other examples of diagnosis include Rett Syndrome and cytomegalovirus (CMV). Nine children did not yet have a confirmed diagnosis. Eight families (11%) reported having more than one child in the family with a disability.

Differences in weekly household income (income after tax, including partner/spouse and excluding allowances e.g. carers allowance etc.) existed across families with some families (n=11) surviving on disposable incomes between €50-€100 or less weekly. The highest proportion of families (N=39) reported weekly disposable household incomes between €251- €750. N=18 families reported disposable income between €751 - €1000 or greater. Only one family in the study did not report disposable household income.

Overall 72% of the mothers and only 5.7% of fathers reported being primary carers for their child. In some cases, care was shared between parents and other family members. Nearly 65% of the primary carer participants gave up employment to care for their child on full time basis at home, with 13% reducing their work hours, which ranged from 15 hours to 3 days per week. 17% of parents did not cease employment and in such cases employed a full-time carer or a family member to provide full-time care for their child. In all situations, there was a loss of household income due to cessation of employment, reducing work hours and the costs of paying a full time carer.

Finally, parents were asked to report whether their child was in receipt of a medical card or not. Table 5.4 presents a detailed description of the three categories of entitlement to free or subsidized health care in Ireland, adopted from (McNamara et al., 2013). Those in category (I) are granted a full medical card which entitles the recipient and his/her dependents to free GP care, prescribed medicines (subject to a per item charge of €2.50 on prescriptions), free access to public inpatient and outpatient hospital services in a public hospital ward (including consultant services), some dental, optical and aural services, entitlement to free maternity and infant care services and a maternity cash grant upon the birth of a child (HSE, 2009). Entitlement to a medical card is decided on the basis of a means test, the income thresholds of which are set nationally and updated annually. However, a medical card can be granted in a situation where the refusal of a medical card would cause undue hardship to people whose income is over the financial guidelines.

Table 5.4 Categories of Entitlement in Republic of Ireland

Categories	GP	Prescription medications	<u>Public hospital care:</u> <u>Acute hospital Inpatient:</u>	<u>Public hospital care:</u> <u>Acute hospital outpatient:</u> (including Emergency Department)
Category I Full medical card	Free	€2.50 per item	Free public care	Free public care
Category I GP visit card	Free	Free above €144.00 out of pocket payment for the month. Free for specific long-term illness conditions.	€75.00 per night subject to annual limit	Free with referral
Category II	About €50 - €55.00. Free Maternity & infant care services	Free above €144.00 out of pocket payment for the month. Free for specific long-term illness conditions.	€75.00 per night subject to annual limit	Free with referral

Having a medical card is an important source of financial support to families in these circumstances. Children are typically prescribed large quantities of medication

that can be expensive to purchase; they are also required to attend several medical appointments. 61 families were in receipt of a medical card, and 9 reported not having a medical card because they were in the process of a long battle to gain access to one.

5.15 Data Analysis

The choice of statistical model to analyse response data is informed by the choice of the error distribution; which in turn, determines whether it is appropriate to use probit, logit or multinomial models for the analysis (de Bekker-Grob et al., 2012). Probit and Logit models are commonly used; in particular, the logit model, as it has a number of appealing features (Hensher et al., 2005). As mentioned in the introductory DCE section, regression results depend on the underlying assumptions on how random components ϵ_{ni} behave in the utility function. We use three distinct models (i) logit (which is similar to probit), (ii) MXL (which relaxes the IIA assumptions) and (iii) Latent class model (which also relaxes the IIA assumptions). The interpretation of Independence of Irrelevant Alternatives (IIA) is that the introduction or removal of a choice has no effect on the proportion of probability assigned to each of the other choices. A concise overview of the models used for the analysis and their respective associated advantages is presented for further clarity in Table 5.5.

Table 5.5: Advantages of using LM, MXL and LCM

Statistical Model	Advantages
Logit Model (LM)	<ul style="list-style-type: none"> • Rudimentary model that can be easily solved. • Most widely used for the analysis of DCE data. • Reports similar results up to scale as Probit model.
Mixed Logit Model (MXL)	<ul style="list-style-type: none"> • Model allows attribute coefficients to vary across respondents and accounts for unobserved preference heterogeneity. • Does not make many assumptions but consequently requires computer simulations to obtain coefficient estimates.
Latent Class Model (LCM)	<ul style="list-style-type: none"> • Assumes there are two or more groups (classes) of respondents with homogenous utility functions. • If data fits LCM with small number of classes, it may have

	more explanatory power than MXL.
--	----------------------------------

5.15.1 (i) The Logit Model

When the error terms are assumed to be independently and identically distributed (with a Gumbel distribution) then we arrive at the logit model (McFadden, 1973). The logit model assumes a logistic distribution of the error terms, whereas the probit model assumes a normal distribution of the error terms. However, the binary probit and binary logit models are expected to give similar results, up to scale (Gerard et al., 2008).

In the logit model, the probability of choosing package *A* is given by

$$P_A = \frac{e^{V_A}}{e^{V_A} + e^{V_B}}$$

It has been claimed (Ryan et al., 2007) that the logit model is inappropriate if the data fails to satisfy a certain criterion i.e. independence from irrelevant alternatives (IIA). This is also known as the Hausman test. Other disadvantages are that it does not incorporate preference heterogeneity and it fails to account for repeated choices by the same individual.

The most basic main effects regression equation to consider is:

$$V = \beta_1 * nnurse + \beta_2 * dnurse + \beta_3 * physio \\ + \beta_4 * speech + \beta_5 * respite + \beta_6 * cash$$

“V” represents the outcome variable, which is followed by the independent variables, *n_nurse* (hours of night nursing care), *d_nurse* (hours of night nursing care), *physio* (hours of physiotherapy care), *speech & language* (hours of speech & language), *respite* (days of respite out of home), and *cash* (a hypothetical cash

maintenance payment). Regressions return estimates of β coefficients and their standard errors. The interpretation of β_3 is the change in utility derived from an additional 1 hour of physiotherapy, and so on.

In Stata, the syntax is:

clogit choice n_nurse d_nurse physio speech respite cash, group(obsid)

A priori we hypothesised, that hours of day nursing care and physiotherapy would be positively related to utility and that out of home respite would exhibit preference heterogeneity within the population.

5.15.1.1 Interactions Effects

Interactions were used to examine differences in preferences based on socio-demographic characteristics i.e., income and geographic location. Parents reported household weekly disposable income after tax in categories ranging from (less than €50.00 per week - €1,501 and greater); we calculated income modes and created two dummy variables, one for “upper” income i.e. any family with income greater than or equal to €625.50 and another for “lower” income i.e. any family with less than or equal to €375.50 disposable weekly income. Parents’ addresses were used to create two dummy variables; “urban” representing families living in urban areas i.e. large town, or city and “rural”, to represent families living in a small town or village. Following this, we created new variables e.g.

*gen byte upperspeech = upper*speech*

*gen byte lowerspeech = lower*speech*

An example regression command was used

*clogit choice n_nurse d_nurse physio upperspeech lowerspeech respite cash,
group(obsid)*

5.15.1.2 Welfare Analysis

The willingness to pay for attribute x_m is typically calculated by

$$WTP(x_m) = -\frac{\partial U / \partial x_m}{\partial U / \partial x_{price}} = -\frac{\beta_m}{\beta_{price}}$$

Since we do not have a price attribute associated with the packages of care, we use as a proxy, willingness to forego payment or willingness to accept compensation for a change in each service instead. Price can convey useful information about the strength of parents' preferences for care services. We use the results to estimate the financial value parents have for particular services:

$$WTP(x_m) \cong \frac{\partial U / \partial x_m}{\partial U / \partial x_{cash}} = \frac{\beta_m}{\beta_{cash}}$$

5.15.1.3 Willingness to Trade

For willingness to accept, the cash payment attribute was used. The same principle applies to any pair of attributes and so we can use this to calculate willingness to trade between any pair of attributes. The lower limit and upper limit were calculated using STATA's inbuilt WTP function.

5.15.1.4 (ii) The Mixed Logit Model

This model allows attribute coefficients to vary across respondents, accounting for unobserved preference heterogeneity and improving realism of model assumptions. In addition, MXL models adjust the standard errors of utility estimates to account for repeated choices by the same individual. Finally, MXL does not assume that the IIA property holds. In Hensher (2001) the model is presented in the form:

$$U_{ni} = V_{ni} + [\eta_{ni} + \epsilon_{ni}]$$

with a deterministic part as before:

$$V_{ni} = \beta_1 x_{1i} + \beta_2 x_{2i} + \dots + \beta_m x_{mi}$$

The difference here is that the error term in square brackets is partitioned into η_{ni} , which is correlated over alternatives and heteroskedastic, and ϵ_{ni} , which is independently and identically distributed over alternatives and individuals.

A disadvantage of the MXL model is that the preference heterogeneity is included in the model by assuming a normal distribution over the coefficients, β . While this may not always be justifiable on theoretical grounds, it is a practical compromise that allows the model to converge.

The proportion of respondent population that has a positive preference for a package attribute x_m is given by the expression below (Kolstad, 2011), where SD_m is the standard deviation of the parameter estimate β_m and $\Phi(*)$ is the normal cumulative distribution function applied to *:

$$Proportion\ Positive(x_m) = \Phi\left(\frac{\beta_m}{SD_m}\right)$$

The Stata syntax applied to night nursing, for example, is:

display normal([Mean]n_nurse/abs([SD]n_nurse))

5.15.1.5 Probabilities of Uptake & Simulations

The simulated uptake estimates the change in probability of picking a package with improved amounts of attribute x_m , relative to a baseline package which has the lowest levels of all attributes as used by (Kolstad, 2011). For a concrete example, consider the change in probability of picking a package with 18 night-nursing hours over one with 12 hours of night nursing, with all other things being equal:

$$P_{nnurse\ 12 \rightarrow 18} = \frac{e^{\beta_{nnurse*18}} - e^{\beta_{nnurse*12}}}{e^{\beta_{nnurse*18}} + e^{\beta_{nnurse*12}}}$$

In Stata, the syntax presented below returns the actual value, standard errors and 95% confidence intervals.

$$\textit{nlcom} (\exp(_b[\textit{nnurse}] * 18) - \exp(_b[\textit{nnurse}] * 12)) /$$

$$(\exp(_b[\textit{nnurse}] * 18) + \exp(_b[\textit{nnurse}] * 12))$$

5.15.2 (iii) The Latent Class Model

The logit model provided useful results, however, the model's assumption of independent irrelevant alternatives (IIA), its failure to encompass preference heterogeneity and its inability to account for panel structure of the data (Greene and Hensher, 2003) means that more alternative models should be used for analysis. Commonly used alternatives include mixed logit models (as presented above) and latent class modeling (LCM). The mixed logit posits a smooth probability distribution across the different respondents whereas the latent class model clusters the respondents into groups/classes, where respondents within the same class have similar utility functions.

The underlying theory of latent class according to Greene and Hensher (2003 p.2) proposes that individual behaviour "depends on observable and on latent heterogeneity that varies with factors that are unobserved by the analyst". Individuals are indirectly organized into a set of classes but it is unknown to the analyst which class contains any particular individual. We can use (LCM) to identify subsets of parents with varying preferences. This will provide useful information for service planning. The probability that individual (q) chooses alternative (i) in a given choice set (t) conditional on falling within class (c) is:

$$P(i, q, t|c) = \frac{\exp(X_{iqt}\beta_c)}{\sum_{j=1}^J \exp(X_{jqt}\beta_c)}$$

This form can be found in (Mentzakis et al., 2011).

The Stata syntax for the LCM assuming two latent classes is given by:

```
llogit choice cash n_nurse d_nurse physio speech respite, id(parentid)
group(obsid) nclasses(2)
```

One can run this for different choices of the number of latent classes. As presented and highlighted in Table 5.4 below, the Consistent Akaike Information Criterion (CAIC) is optimised for two latent classes.

Table 5.6 Consistent Akaike Information Criterion (CAIC)

Classes	LLF	Nparam	AIC	CAIC	BIC
2	- 458.21	13	942.43	984.47	971.47
3	-430.54	20	901.09	965.77	945.77
4	-414.24	27	882.48	969.80	942.80
5	-403.87	34	875.74	985.70	951.70

5.16 Results

Table 5.5 presents the results of the (main effects) logit (fixed effects) model with interactions. Given the coding in Table 5.2, the positive and significant coefficients for “day nurse”, “physiotherapy”, “speech & language therapy” and “cash maintenance payment” all indicate that parents have a preference for more of those services and are more likely to choose a package of care with those particular service attributes in it. These coefficients were significant and in the expected direction. The large p-value on the “night nursing” coefficient indicates a non-uniform (heterogeneous) attitude toward night nursing care and, finally. “Out of home respite” had both a negative coefficient and an insignificant p-value. This suggests that families are either receiving enough of this service already or are indifferent about receiving more of this service. These findings were in line with expectations and support the theoretical validity of the model. We also interacted

both income and location with all service types to examine socio-demographic differences in preferences. Lower income families showed particular preferences for day nursing care, physiotherapy and a cash maintenance payment (although the cash maintenance payment had a small coefficient). Evidence from the UK found that it costs £12,000 to raise a child with a disability in comparison to non-disabled child (£7,355) from birth to 17 years (Dobson and Middleton, 1998a). According to another UK study by Beresford (1994) one of the greatest areas of unmet need is financial support to help with the additional costs of caring for a child with a disability. Upper income families showed preference for day nursing care, physiotherapy and speech and language therapy. Families living in urban areas had a preference for physiotherapy and both night nursing and day nursing care. This finding may reflect that families living in urban areas live long distances from other family supports, so they have a preference for both night and day nursing care to serve as a replacement for a lack of informal support. Families living in rural areas had a preference for speech and language therapy, day nursing care and physiotherapy, reflecting perhaps a shortage in some under-resourced rural locations. The results indicate some differences in preferences based on socioeconomic characteristics; however, with such a small sample size it is difficult to make any inferences.

5.16.1 (i) Logit Model Results

Table 5.7 Multinomial Logit Model with Interactions

Coef	β	SE	<i>p</i>-val
n_nurse	.0087	.0065	0.178
n_nurseupper	.0041	.0090	0.642
n_nurselower	.0132	.0089	0.137
n_nurseurban	.0384	.0119	0.001
n_nurserural	-.0015	.0077	0.841
d_nurse	.0196	.0022	0.000

Coef	β	SE	<i>p</i>-val
d_nurseupper	.0233	.0030	0.000
d_nurselower	.0163	.0028	0.000
d_nurseurban	.0122	.0035	0.001
d_nurserural	.0221	.0026	0.000
Physio	.4470	.0505	0.000
Physioupper	.4070	.0694	0.000
Physiolower	.4870	.0701	0.000
Physiourban	.4256	.0900	0.000
Physiorural	.4452	.0602	0.000
Speech	.1982	.0534	0.000
Speechupper	.2097	.0752	0.005
Speechlower	.1871	.0739	0.011
Speechurban	.2023	.0975	0.038
Speechrural	.1808	.0640	0.005
Respite	-.0484	.0498	0.331
Respiteupper	-.0496	.0702	0.479
Respitelower	-.0471	.0692	0.496
Respiteurban	-.0942	.0914	0.302
Respiterural	-.0326	.0599	0.586
Cash	.0030	.0010	0.005
Lowercash	.0038	.0014	0.007
Uppercash	.002	.0014	0.127
Log Likelihood	-489.0		
LR chi2(6)	169.0		

<u>Coef</u>	<u>β</u>	<u>SE</u>	<u>p-val</u>
Prob > chi2 (6)	0.000		
Pseudo R2	0.147		
Number of individuals	69		
Number of observations	1656		

5.16.1.1 Welfare Analysis

Table 5.8 below presents the results of how much financial compensation parents would accept to forgo a particular service attribute.¹

Table 5.8 Welfare Analyses

	WTP	95% CI
N_nurse	€2.92 X 8hrs = €23.20	(€7.20 - €1.35)
D_nurse	€6.54 X 8hrs = €52.32	(€10.6 - €2.39)
Physio	€148 = 1hr	(€258 - €39)

¹ Typically in economics, we are looking to estimate willingness to pay (WTP) for a particular good or service but in this case we look for willingness to accept compensation (WTAC) for a loss of a particular good or service.

Speech	€66 = 1hr	(€124 – €6.9)
---------------	-----------	---------------

The value reported for physiotherapy gives us a clear indication about the importance of getting more of that service. Using coefficient estimates from Table 5.5, we show that on average, parents would need to be compensated by the sum of €148.00 ($.4470 / .0030 = 148.00$) for a loss of one hour of physiotherapy, which is almost 3 times the original cost of a physiotherapy appointment. All other things being equal, this is a clear indication of parents' needs for additional physiotherapy. A similar albeit, smaller amount of €66.00 is the required compensation to forgo one hour of speech & language therapy. Both results are confirmed by the fieldwork findings where parents expressed concerns over a shortage of physiotherapy and speech & language therapy. Day nursing care exhibits similar values where parents would need to be compensated by €52.32 for a loss of eight hours of day nursing care. Parents have substantially lower compensation requirements (€23.20) for one shift (eight hours) of night nursing care, which may suggest this service is sufficiently supplied. Out of home respite was omitted from the analysis, as it was not statistically significant.

To conclude, the WTAC analysis tells us that physiotherapy is the most valued service, in a monthly package of care, for parents caring for a child with a life-limiting disability. Other services that parents would like more of are speech & language therapy, day nursing care and night nursing care. Out of home respite consistently appears to be the least important service to parents.

5.16.1.2 Willingness to trade between services:

As previously stated, price can convey useful information about the strength of parents' preferences for care services. Table 5.9 below presents the results of parents' willingness to trade between services. An example of how one interprets the results is where parent's are willing to trade €65.96 units (euros) of cash for 1

unit (hour) of speech therapy. In the other direction, they are willing to trade 0.01516 of an hour of speech therapy for 1 euro. ($1/65.96 = 0.01516$).

Table 5.9 Willingness to Trade

	n_nurse	d_nurse	physio	speech	respite	cash
n_nurse	1	2.239 (-0.94,5.42)	50.90 (-24.9,126.7)	22.57 (-14.2,59.34)	-5.511 (-19,8.01)	0.3422 (-0.16,0.84)
d_nurse	0.4467 (-0.19,1.08)	1	22.74 (16.45,29.02)	10.084 (4.26,15.9)	-2.462 (-7.5,2.57)	0.1529 (.06,0.25)
physio	0.01965 (-0.01,0.04)	0.04398 (0.31,0.56)	1	0.4435 (0.20,0.69)	-0.1083 (-0.32,0.11)	0.006723 (0.002,0.012)
speech	0.04431 (-0.028,0.12)	0.09917 (0.04,0.16)	2.255 (1.02,3.5)	1	-0.2441 (-0.74,0.25)	0.01516 (0.002,0.029)
respite	-0.1815 (-0.63,0.26)	-0.4062 (-1.23,0.424)	-9.236 (-28,9.6)	-4.096 (-12.4,4.24)	1	-0.0621 (-0.18,0.062)
cash	2.922 (-1.36,7.2)	6.54 (2.4,10.67)	148.74 (39,259)	65.96 (6.95,125)	-16.104 (-49,17)	1

5.16.2 (ii) Mixed Logit Results

The results from the mixed logit analysis with 500 halton draws is presented below in Table 5.10. As in the previous logistic model results, preference heterogeneity for out of home respite is revealed as a large P value. The coefficient for night nursing is only significant at the 10% level, whereas all remaining coefficients are significant at the 5% level excluding out of home respite care. Physiotherapy is once again seen to be the most valued service, with second-placed speech and language having a coefficient roughly half as large. Every additional hour of day nursing is seen to be about 30% more useful than every extra hour of night nursing.

Table 5.10 Mixed Logit Model

Choice	Coef.	Std. Err.	P>z
Mean			
Cash	.0053	.0017	0.002
n_nurse	.0346	.0192	0.072
d_nurse	.0455	.0080	0.000
physio	1.013	.1698	0.000
speech	.4557	.1000	0.000
respite	-.1237	.1471	0.400
SD			
n_nurse	.1236	.0226	0.000
d_nurse	.0491	.0091	0.000
physio	.9106	.1682	0.000
Speech	.2403	.2034	0.237
Respite	1.034	.1840	0.000
Number of	1656		

Choice	Coef.	Std. Err.	P>z
obs			
LR chi2(5)	146.42		
Log likelihood	-415.83		
Prob > chi2	0.0000		

5.16.3 Probabilities of selecting a package based on attributes

Using mixed logit estimates, we calculate probabilities in selecting a package based on its attribute. See note ²

Table 5.11 Probabilities

Choice	b	z	P> z	e ^b
n_nurse	0.008	1.348	0.178	1.0088
d_nurse	0.019	8.725	0.000	1.0199
physio	0.447	8.852	0.000	1.5637
speech	0.198	3.711	0.000	1.2193
respite	-0.048	-0.972	0.331	0.9527
cash	0.003	2.792	0.005	1.0030

Table 5.11 presents the odds ratios (OR) of picking a particular package that has an additional amount of a service offered. Parents are more likely to pick a package of care that has an additional hour of physiotherapy by a factor of 45% (0.44708).

² * Note: A rough approximation of an allocation of day nursing would typically be 8 hours. We use the d_nurse coefficient $0.019 \times 8 = 0.152$ which would be comparable with speech = 0.198.

Speech and language therapy by a factor of 20% (0.19827), day nursing care 15% (see note 2) and the cash payment has little to no effect. There is no significant effect for respite care or night nursing.

5.16.4 Simulations and Predicted Uptake

Table 5.12 presents the probabilities of choosing a given package of care when the levels of attributes are changed. We consider the change in probability of taking a baseline (lowest service level) due to a change of the level in one of the service attributes.

The probability of choosing a package increases by 21.8% if day nursing increases from 30 hours to 50 hours in a month. The probability is even higher (49%) when increased to 80 hours. From an initial package of care with one hour of physiotherapy, an alternate package with two additional hours is twice as likely to be chosen as a package with one hour and these results are similar for speech and language therapy. The cash maintenance payment is also valued and the probability of selecting a package, with an increase of €50 - €100, is 7.5%; an increase to €150.00 means it doubles to 14%. There was no significant effect on night nursing care or out of home respite care.

5.16.4.1 Simulations

Table 5.12 (Simulations) Effect of Changes in Levels of Attributes

Choice	Uptake	SE	P val
Nnurse 16-24hrs	.0351(3.5%)	.0260	0.177
nnurse 16-36 hrs	.0876(8.7%)	.0646	0.175
dnurse 30-50 hrs	.2186(21.8%)	.0289	0.000
dnurse 30-80 hrs	.4928(49%)	.0516	0.000
Physio 1-2 hrs	.2198(21.9%)	.0240	0.000
Physio 1-3 hrs	.4194(41.9%)	.0416	0.000

Choice	Uptake	SE	P val
Speech 1-2 hrs	.0988(9.8%)	.0264	0.000
Speech 1-3 hrs	.1957(19.5%)	.0513	0.000
Respite 1-2 hrs	-.0241(2.4%)	.0248	0.331
Respite 1-3 hrs	-.0483(4.8%)	.0497	0.330
Cash 50-100	.0750(7.5%)	.0267	0.005
Cash 50-150	.1491(14.9%)	.0526	0.005

5.16.5 (iii) Latent Class Results

The LCM presented in Table 5.13 identified two classes of parents with different preferences for care services. The first class of parents showed a preference for more day nursing care, physiotherapy, speech and language therapy and out of home respite care. Although out of home respite care was only significant at the 10% level, this does provide evidence of theoretical validity i.e. the prior assumptions of differing preferences for out of home respite care. The second class of parents had a preference for more night nursing care and physiotherapy in a given monthly package of care.

Physiotherapy was the only attribute that had both a positive and significant coefficient in both classes, indicating perhaps the need for more physiotherapy for children with LLC's. Research on the service needs of UK families found that one of the greatest areas of unmet need was in the area of child rehabilitative services for improving mobility and communication. In particular, parents perceived a shortfall in the availability of physiotherapy and speech and language therapy (Sloper and Turner, 1992). The findings from the latent class analysis demonstrate heterogeneous preferences amongst parents caring for a child at home. The class share reported was however (0.798 vs. 0.202) where class 1 made up approximately 80% of the population so the size of each class is quite different. Nonetheless, the results obviate the need for more rehabilitative services for children with LLC's.

Table 5.13 Latent Class Model

	Class 1	S.E	p-value	Class 2	S.E	p-value
Cash	.0028	.0012	0.023	.0094	.0042	0.025
N_nurse	.0161	.0081	0.048	.1493	.0393	0.000
D_nurse	.0268	.0028	0.000	-.0067	.0062	0.282
Physio	.5229	.0606	0.000	.4516	.1724	0.009
Speech	.2530	.0627	0.000	.3476	.2206	0.115
Respite	-.0997	.0612	0.104	.1191	.1668	0.475

5.17 Summary Discussion

Chapter 5 is a first attempt to present empirical evidence regarding parents' preferences of care services for children with life-limiting neurodevelopmental conditions in Ireland. The study identified several important service attributes that parents would like more of in relation to meeting the needs of their child. Preferences for more of the following services were identified: physiotherapy, speech & language therapy, day nursing care and a cash maintenance payment. The findings provide important information on what is needed on a regular basis for these children to thrive. The three statistical models utilised in this study (the logit, mixed logit and latent class models) provide unique insight, in particular the latent class model where two distinct groups of parents with heterogeneous preferences were identified. In addition, using the estimates from the three models allowed us to calculate predicted uptake, which can be used to inform policy makers on the development of care services for this population. The WTAC estimates provide a monetary valuation for particular service types, the results of which, demonstrate further evidence on parents' valuation of care services.

This study provides evidence of unmet need in relation to physiotherapy and speech & language services for children with LLC's. The qualitative study (presented in chapter 4) revealed several issues around rehabilitative services such as; poor access, poor availability, and disparities between parents and professionals

views. The findings from the DCE analysis obviate the need for more of these kinds of services. When all other things are considered, parents are willing to give up a lot to obtain more of these services. Parents may benefit from having more flexibility in the use of the financial grant they receive from the JJF. This would enable them to privately finance additional services. Although this is not ideal, some parents do privately finance these services, as there is a shortage of free services available. The cash maintenance payment offered in the experiment was valued more by parents in the lower income categories and although it is a hypothetical service, it is needed by this particular group of families to help with the additional costs of caring. Chapter six presents the results of section two of the quantitative questionnaire on the out of pocket costs of caring and provides important context for the findings of the discrete choice experiment.

These findings are not only important for care providers but are essential for planning both sustainable and satisfactory care models in the future. The results are especially useful in the disability health-care setting where resources are highly constrained by recent budget reductions and poor statutory funding. The DCE results can be used to inform the implementation of the recently published national guidance on care for these children in Ireland (Laura Lynn, 2013). The voluntary and statutory health care providers can also use these findings to ensure a more patient focused service model is implemented in practice. A very useful extension of this work would be to further explore the reasons for parents' heterogeneous preferences and to examine the preferences of health care professionals.

5.18 Strengths and Limitations of the Study

Results from these models provide estimates of the value and importance to respondents but do not often predict future behaviour or health outcomes according to Bridges et al. (2011), therefore, a number of limitations are acknowledged for this study. The sample size (n=69) was large enough to produce significant results but too small to make sub-group analysis generalisable to the population. The diverse range of life-limiting conditions experienced by these children and the uncertain nature of such conditions means that care needs are likely to vary throughout illness trajectories; that is, one model does not fit all. It would therefore be beneficial to

repeat this study at another time to reflect altering needs but also incorporate the views of health care professionals. In addition, DCE's are known to be cognitively demanding (Ryan et al., 2007) and this was a concern for this study, however, only 2% of respondents reported finding the task "extremely difficult", which was promising. Also, like other DCE studies, we used a main effects design, which assumes that the impact of one attribute on utility is independent of the other attributes in the choice scenario but this may not be true; one study suggests using two-higher order interactions to deal with this issue but this was outside the scope of this study (Naik-Panvelkar et al., 2012).

This study did not include an opt-out option, and if this was incorrectly excluded, an overestimation of the values obtained may occur (Morey et al., 1993), however, we were confident that an opt-out would not represent a realistic situation for parents as they are in a no choice situation. In addition, the restricted set of attributes and levels limits realism and applicability particularly so for the case when estimating WTP/WTA, where the levels of cash maintenance payments were chosen by the researchers which in turn influences the WTP estimates (Ratcliffe, 2000) so should be treated with caution.

In this chapter, the findings have been reported for parents' preferences of care services. A detailed discussion on these findings will be presented in Chapter 7.

Chapter 6

Evidence on Financial Impacts of caring

6.1 Introduction

The purpose of chapter 6 is to present the results of a descriptive study on the financial costs of caring for a child with a life-limiting condition at home in the Republic of Ireland. Section two of the DCE questionnaire (presented in Chapter 5) asked families to report their household income and out of pocket costs in relation to providing care. This chapter is an extension of the work presented in Chapter 5 and serves to provide further clarity and context to the findings of the qualitative and quantitative studies from chapters 4 and 5.

6.2 The Economic Costs of Caring

6.2.1 Introduction

Families caring for children with disabilities are characterized by “financial hardship, stress and anxiety as a result of social barriers, prejudices and poorly conceived service provision” (Dowling and Dolan, 2001 p.25). Children with life-limiting conditions have exceptional needs due to the nature of their disabilities and as a result, families caring for these children face a higher risk of experiencing poverty and debt (Meyers et al., 1998, Copps et al., 2007, Blackburn et al., 2010, Martin et al., 1998). According to a recent UK cost survey (Family, 2014), these families are more likely to go without food and heating and, in some cases, require financial loans to pay for basic goods. In many cases, the additional costs are borne by families and, “when poverty and disabilities intersect, the public and private costs associated with children’s care have important policy implications” (Meyers et al., 1998 p.210). Measuring the extent of the financial burden is not only important for the identification of support needs, it also has public policy relevance.

The literature review in Chapter 2 provides an overview of the costs associated with caring for a child with a disability. Research gaps were evident in relation to the direct and indirect costs borne by families, particularly for this population group and existing research is in most cases dated (Joseph Rowntree Foundation, 1999, Meyers et al., 1998, Anderson et al., 2007, Glendinning et al., 2001). In Ireland, to date, research on the costs of disability has focused on the working age population (Cullinan et al., 2011). In contrast, our baseline study was the first comparison of costs on the provision of care for children with LLC's in the acute hospital setting versus the home care setting in Ireland (Revill et al., 2013). The descriptive study presented here adds a contribution to the literature on the direct and indirect financial costs of caring.

Results from the qualitative study (reported in chapter 4) revealed some of the financial challenges parents were experiencing as a result of caring for their child. Consequently questions were added to section two (Appendix 13) of the quantitative questionnaire to obtain a more detailed insight into the financial costs of caring, specifically exploring the additional direct and indirect costs. The chapter commences with a brief overview of the financial support mechanisms available to families in Ireland and a description of the data and methodology used for the study. Subsequently, the results of the study are outlined and a brief discussion on the findings and the study limitations are outlined.

6.2.2 Aim of This Study

Section two of the quantitative questionnaire (reported in Chapter 5) asked additional questions on the direct and indirect economic costs of caring. The purpose of asking such questions was to examine the financial costs of caring for a child with a neurodevelopmental life-limiting condition at home. We adopted the following definitions;

- A direct cost is defined as expenditure required in order to meet the needs arising from the disability. Examples include, medication, travel costs, electricity for a technology dependent child.

- Indirect costs are defined as a reduction in financial wealth in comparison to a non-disabled person as a result of the disability e.g., having to give up work to care for your child, which results in a loss of household income.

As already outlined in chapter 5, the DCE questionnaire gathered a response from sixty-nine parents across the Republic of Ireland. The analysis of the results of section two of the questionnaire reveal the significant out of pocket costs associated with caring for a child with a LLC.

6.2.3 Methodology

Our conceptual model integrates income from a broader perspective, including income from social policies and employment income. The type of ‘costs data’ to be collected in this study was influenced by the classifications of the costs related to disability by Berthoud (1991). An important concept to consider is *need* versus *cost*, where extra costs are incurred as a result of the extra needs that disabled people require. These costs include:

1. **Special:** Goods and services that disabled people need but non-disabled people do not need. An example would be medication, special equipment, like a car seat or chair to help support a child. Other examples include special needs toys and clothing.
2. **Additional:** Also known as “disability-created”. These are items every person needs but disabled people need more of. An example would be petrol costs and other replacement costs like washing machines.
3. **One-off:** An item that only needs to be purchased once, an example of which would be adaptations to the disabled persons i.e. making the home wheelchair accessible or making adaptations to the car.
4. **Recurrent/Intermittent:** In the case for children with LLC’s, this would include nursing care, a carer or home help.

6.2.4 Survey Question

In section 2 of the quantitative DCE questionnaire, we presented parents with the following question in relation to the additional costs of caring for their child:

- *We understand caring for a child with complex care needs has many additional financial costs. We have listed some examples below. Please fill in approximate costs and other examples not listed below. Please also include non-reimbursed expenses (yet to be paid)*

Table 6.1 presents a list of items under the remit of the four cost classifications outlined above. Parents were asked to report the costs associated to each item listed and were encouraged to add any additional items not listed and were provided with extra space to do so.

Table 6.1 Parents were asked to report the costs

Service Type:	Cost:
Private Physiotherapy / other services	
Equipment for your child	
Travel costs (fuel + accommodation + babysitting costs)	
Electricity bills / oil bill / heating bill	
Phone bill	
Medication costs	
Adaptations to home / car	
Non reimbursed expenses (yet to be paid):	

6.2.5 Overview of Financial Mechanisms Aimed at Supporting Families in Ireland

The following supports are intended to offset, at least partially, the additional costs incurred by families caring for a disabled person. The families in this study reported if they were in receipt of the following:

- **Carer's Allowance:** If parents find themselves under a specific income threshold and are caring for a person who needs support, because of age or a physical/learning disability or illness, they can apply for a carer's allowance. To avail of this grant, parents must be in the position to care for this person on a full time basis. The weekly payment starts at €204.00 and increases depending on the number of people being cared for (Carer's Allowance, 2014). In this study, 38/69 families were in receipt of the carers allowance, leaving 31 families without this payment. Families not in receipt of the carers allowance either had incomes above the required threshold or were actively pursuing the allowance at the time this study was conducted.
- **Annual Respite Grant:** The respite care grant is provided to families by the department of social protection to help finance additional support; for example, paying for respite care services or additional care needs for their child. A payment of €1,375 is paid to carers annually. All families in this study (N=69) were in receipt of this grant.
- **Domiciliary Care Allowance (DCA):** If you are caring for a child who has a severe disability and requires substantial care and attention, in comparison to a non-disabled child of the same age, then parents are entitled to a (DCA) payment. The monthly payment is €309.50 per child with a disability (Health Service Executive, 2013). N=69 families were in receipt of the DCA grant.
- **Medical Card:** Chapter 5 presents a detailed overview of the three categories of entitlement to free or subsidized health care in Ireland. Those in category (I) are granted a full medical card, which entitles the card holder and dependents to free GP care, prescribed medicines (subject to a per item charge of €2.50 on prescriptions with a limit of €25.00), free access to public inpatient and outpatient hospital services in a public hospital ward

(including consultant services), some dental, optical and aural services. Entitlement to a medical card is decided on the basis of a means test, the income thresholds of which are set nationally and updated annually. However, a medical card can be granted in a situation where the refusal of a medical card would cause undue hardship to people whose income is over the financial guidelines. 8/69 families reported not having a medical card at the time of this study.

An important consideration in the context of financial supports in Ireland is that disability service budgets were reduced by 1.25% in 2013 (€1,554 million to €1,535 million). Although a small reduction, citizens caring for disabled people in the community may face additional financial strain as a result. Parents of children with complex disabilities experienced additional challenges when the government announced a 19% reduction in the annual respite grant (€1,700 to €1,375) in 2012's budget. In 2012, there was a €10.00 reduction in the monthly child benefit allowance, which reduced it from €140.00 to €130.00 for the first three children and €140 thereafter. All of these cuts place additional stress on families that are already in difficult financial circumstances.

6.3 Results

Sixty-Nine families responded to the questionnaire (already reported in chapter 5) and completed the 'Economic Situation' in Section 2. The sections were fully completed, with the exception of one family, who did not report total household income after tax. Nine families did not report loss of income. Otherwise, clear indications of costs were provided. Families reported the additional costs in monthly and yearly amounts; yearly amounts were divided by twelve to calculate monthly costs. Table 6.2 below presents an overview of the results of the costs survey. Annual average costs were calculated for each domain (as presented in Table 6.2 below) in the following way: For example: *special costs*: $N=36/69$, i.e., 52% of families reported nonzero annual rehabilitation costs (physiotherapy, occupational therapy and speech and language therapy). Averaged over $N=69$ families, the special costs amounted to €1,143 annually. The total cost burden is

calculated as the total of *special costs*, *additional costs* and *loss of income* minus the *financial support* received.

Table 6.2 Costs associated with caring for a child with a complex disability

Costs	Families N = 69	Mean Annual Amount
<i>Special costs</i>		
Physiotherapy / OT / Speech	36(52%)	€1,143
Medication Costs	36(52%)	€367.00
Travel Costs Fuel / Parking	55(79.7%)	<u>€2,692</u>
		€4,202 (A)
<i>Additional costs</i>		
Electricity / Oil / Heating	52(69%)	€1,048
Phone Bill	52(75%)	<u>€660</u>
		€1,708 (B)
<i>Loss of income</i>		
Loss of Income	48 (70.5%)	€23,871
No loss of Income	12 (17.3%)	0.00
Missing	9 (12.5%)	0.00
		<u>€23,871 (C)</u>
<i>Financial support</i>		
Carers allowance	38 (55.8%)	€9,702
Annual Respite Grant	69 (100%)	€1,375
Annual DCA	69 (100%)	<u>€3,714</u>
Avg Annual Financial Support		<u>-€14,791 (D)</u>
<i>Avg Cost Burden (A+B+C+D)</i>		€14,990
HH Income After Tax	68 (98.5%)	€28,000
One-off Payments		

Costs	Families N = 69	Mean Annual Amount
Adaptations to Home / Car	36(52%)	€7,329
Equipment	39(57%)	€353.70
Other	25(36%)	<u>€693.90</u>
		€4,275

6.3.1 The Direct and Indirect Costs of Providing Care to Children with LLC's:

6.3.1.1 Direct costs

Special Costs

- Physiotherapy OT & Speech:** 36/69 families reported paying additional out of pocket costs for physiotherapy, occupational therapy and speech and language therapy. The average monthly payment was €95.30, which equates to €1,143 annually. These figures range from €50.00 - €520.00 monthly which may reflect affordability. Twenty-three families reported paying zero out of pocket for additional services and ten families did not provide any information on these costs.
- Medication Costs:** 36/69 families reported paying an average of €30.60 on medication costs every month, which equates to €367.00 euro annually. These payments range from €10.00 - €216.00. The remaining twenty-three families did not pay any out of pocket costs towards medication. Some of these costs are in relation to items that are not covered by the medical card. Parents who are in receipt of a medical card are only required to pay up to a maximum of €25.00 euro for prescription items. The remaining twenty-three families did not report any medication costs and again ten families did not report any information on costs.

- **Travel Costs - Fuel / Parking:** 55/69 families reported paying €224.40, which equates to €2,692 annual costs. Monthly amounts ranged from €30.00 to €1,000 euro a month. Typical costs include fuel costs, parking costs and paying for a babysitter to care for other children while attending hospital appointments. The remaining fourteen families did not report any travel costs.

Additional Costs

- **Electricity Bills, Oil and Heating:** 52/69 families reported paying a monthly average amount of €174.00 for electricity bills, which amounts to €1,048 annually. This amount ranges from €30.00 to €800.00 euro a month. Typically, technology dependent children require equipment to be charged on a full time basis and heat and lights needs to be switched on 24/7, especially when nurses/carers are providing care through the night. The lower costs may reflect families who do not have a technology dependent child. The remaining seventeen families did not report any electricity costs associated with their child.
- **Phone Bill:** 52/69 families reported a monthly average of €55.00 euro for phone bill costs, which equates to an annual cost of €660.00. The remaining seventeen families did not report phone bill costs.

One off Payments

- **Adaptions to Home & Car:** 36/69 parents reported adaptations costs for their home or car. These costs can occur intermittently over a child's lifetime and can vary significantly; for example, in one case, a car seat cost €2,220, while a house adaption to make it wheelchair accessible cost €25,000. Thirty-three families did not report any costs in relation to home adaptations.
- **Equipment:** 39/69 parents are required to buy special clothing or equipment for their children such as sensory lights that cost up to €1,000. Many parents feel the pressure to buy everything suggested to them because it makes them

feel they are doing their best for their child. Thirty families did not report any additional equipment costs.

- **Other:** 23/69 parents have an average annual cost of €693.90 to purchase specific other items in relation to caring for their child. One example provided was for support shoes that cost €300.00. Other items of clothing include pressure clothing, extra jackets, extra bibs, TheraTogs and private physiotherapy. One mother reported paying €500 in the previous year on special clothing costs. Forty-six families did not report any other costs in relation to caring for their child.

The items and costs listed above are similar to recent cost estimates in the UK (Counting the Costs of Disability, 2014) for disabled children in comparison to non-disabled children in the UK. For example the cost of a car seat for a child with a disability is £600 in comparison to £149.00 for a non-disabled child. A sensory wooden toy is £1,000 in comparison to £16.99 for wooden blocks for a non-disabled child.

6.3.1.2 Indirect Costs

- **Loss of Income** 48/69 families reported an average annual loss of income of €23,871. This ranged from €2,000 to €113,000. Only one father ceased employment full time in order to care for their child, showing that mothers are more likely to give up employment in these situations. Twelve parents did not report a loss of income as they were either made redundant or unemployed prior to having their child. Nine families did not report their status.

Total Cost Burden on Families

On average families caring for a child with a life-limiting condition in Ireland are up to €14,990 worse off annually, that is 53% of their household income is allocated to the additional costs of caring. In estimating this figure, we considered the costs presented in Table 6.1 above. This number excludes one-off/intermittent payments which may underestimate the cost burden. We performed a coarse sub-

group analysis where we partitioned families into those that had less than €500 disposable household income (N=35) and those with greater than €500 household income (N=33). The differences in average out of pocket spending were negligible for all the different costs categories. This suggests that there is a disproportionate burden on families with less disposable income. A recent study on out of pocket payments for childhood stroke by Plumb et al. (2015) found that parents from different financial strata encounter similar out of pocket costs when a child develops a neurological illness that requires chronic care and rehabilitation. A key finding of this present study is the “hidden costs” identified such as; electricity and heating bills, home adaptations and the costs of special needs toys and equipment which are all necessary for the child’s ongoing care. Devoting such a high proportion of income to the health care expenses is defined as catastrophic health expenditure (CHE), where families are forced into poverty as a result of paying high out of pocket payments (Marmot et al., 2010). The families in this study devote on average up to 50% of their household disposable income towards the costs of care and as a result face a higher risk of experiencing poverty. However, a limitation of this study is that the results reported use averages only so the generalisability of these results warrant caution. A recent study by Kronenberg and Barros (2014) on the drivers and protection of catastrophic health expenditure in Portugal recommended exemption of co-payments for vulnerable populations, particularly children and elderly populations with disabilities. Only 50% of the families in this study were in receipt of the carers allowance and nine families reported not having a medical card which are concerning statistics for this small sample. The results of this study provides evidence that children with life-limiting neurodevelopmental conditions represent an unrecognised cost to families and society in Ireland. A recommendation of this study would be to revise existing financial support mechanisms as current supports are failing to adequately meet the needs of this population.

6.4 Conclusion

The findings from section two of the DCE questionnaire clearly reveals families face significant additional out of pocket costs in relation to caring for their child and lose, on average, €14,990 when all other income supports are considered. High cost

items reported by families include fuel costs, parking costs, special needs toys, clothing and equipment, and finally, the substantial adaptations to home and car that is required to accommodate their child's needs. The predominantly single income families face the additional challenges of no options to return to work given the seriousness of their child's illness. A large number of mothers gave-up full time employment to care for their child which resulted in an average loss of household income of €23,871 and although this figure varies widely, it still represents a large loss for families. Although this study represents a small sample of families in Ireland, it still presents evidence on the significant financial impact of caring for a child with a life-limiting condition in Ireland. Another extension of this research may be to investigate how families finance their health expenditure, for example, through, savings, borrowing or selling existing assets. Children with LLC's are surviving longer than before and this present study only covers the age range of 1-7 years, the question remains, are the out of pocket experienced by these families sustainable in the long term? The survival rates of this vulnerable population is increasing and current policy needs to be addressed to support and maintain the future provision of care at home.

6.5 Further Commentary Provided by Parents

The formal analysis of the qualitative and quantitative studies revealed important findings on parents' experiences and preferences of care services. At the end of the DCE questionnaire, parents were afforded the opportunity to provide comments on pressing issues. In all cases (N=69), parents chose to avail of this opportunity, in some cases making extensive comments. The most common issues mentioned were in relation to poor access to rehabilitative services, uncertainty about future service provision, financial concerns and a need for more integration between services. A sample of these comments is provided in Appendix 16. This commentary enhances the richness and further validates the overall findings of this study.

6.5.1 Counselling Services / Emotional Support Services for Parents

Throughout the qualitative phase of the study (reported in Chapter 4), most parents reported a need for emotional support services. Additional questions were asked about families' need for counselling services, which is reported below. Three questions were added to section 2 (Appendix 13) of the quantitative questionnaire in relation to counselling services for parents:

1. Do you think a counselling service should be offered to parents as part of a care package?
2. If you had to pay for a counselling service, how much would you be willing to pay for this service?
3. If offered, would you use 2 hours of your Jack & Jill Foundation allowance to pay for a counselling service for you or your partner?

In their answers, 51/69 families would like to have counselling services offered as part of a care package. Roughly half (33/69) would pay an average of €30.00 for a counselling service; the remaining families said they could not afford to pay for this service and that they would find it difficult to find a babysitter to cover while they attended the service. Finally, 38/69 families said they would use two hours of their Jack & Jill Children's Foundation allowance to pay for counselling services; the remaining families said they would not. It is interesting to see that the majority of the families would like to have some form of emotional support and the need for this services is reflected in their willingness to use their JJF funding more flexibly. This is an important finding, particularly in relation to the findings of this study. This study highlights the varying challenges parents experience be it, the intensity of caring for a sick child to a shortage of services directly related to their child, this is compounded by the loss of essential financial support mechanisms and high costs of providing care. It is no surprise, therefore, that families are in need of interventions to help them cope with these extremely challenging situations.

6.6 Discussion of Strengths and Limitations

This descriptive costs study relies on estimates from a small sample (N=69) of families caring for children with life-limiting conditions in Ireland, but it nonetheless provides important insight into the financial impacts of caring for children with LLC's. It would be useful to conduct a more comprehensive study to identify the larger scale impact of out of pocket costs on families. The results of this costing study provides an important backdrop from which to draw further conclusions on the preferences of care study in chapter 5. Understanding the extent to which families are affected financially allows us to further understand the rationale behind choosing the services they did. The results of this study may not be generalizable to other families caring for children with less severe disabilities in Ireland, but this study provides evidence from the perspective of a very under-researched population and has important public policy implications.

Chapter 7

Discussion of the Findings

7.1 Introduction

The aim of this study was to explore parents' experiences in relation to services for their child, identify service preferences and examine out of pocket costs for families caring for children with life-limiting neurodevelopmental conditions in Ireland. The focus of this chapter is to discuss the overall findings of this study. Firstly, the results from phase one of the study is discussed in relation to parents' service experiences, this is followed by a discussion on phase two's findings from the quantitative study on parents' preferences of care. The findings from the descriptive study, on the financial impact on families, are integrated throughout the discussion chapter, which is congruent with the mixed methodological design chosen for this study.

7.2 Phase 1: Parents' Experiences of Caring for Children with Life-limiting Disabilities at Home

7.2.1 Transitioning from Hospital to Home

The findings from this study reveal the transition from hospital to home to be the most vulnerable time for parents, where they felt isolated and disconnected from key supports which is similar to findings by Carnevale et al. (2006). The management of extremely rare and complex conditions in children is frequently left to parents to manage alone which was also reported by Rahi et al. (2004). Brinchmann (1999) found that parents compared their homes to prison because of the dependency of their child and their intensive care requirements. The parents in this study described being at breaking point before finally receiving the services

their child needed, which in some cases, only arose from the outcome of a major crisis. These findings are similar to a recent UK study by a voluntary organization, “Action for Children” (Action for Children, 2013), who examined the impact of government spending decisions on children with disabilities. They found that service infrastructures were collapsing, with a loss of early intervention work, which meant that as a result, services were moving towards dealing with families in crisis situations only.

Parents in this study frequently reported the following issues: poor service set-up in relation to referral to essential supports, extensive delays in receiving services and a lack of information available which is congruent with Heaton et al. (1999) findings on parents experience of transition from hospital to home. Discharge management, and the need to improve the coordination of care between hospital and community services, has been raised a key concern in Ireland’s national guidelines and needs assessment reports (Laura Lynn, 2013, DOHC 2005, DOHC 2010). The need for early referral and coordinated care was exemplified by two parents’ accounts of the very positive transitions they experienced, as a result of strong communication between hospital and community services as found by Appleton et al. (1997) and Turchi et al. (2014).

Coordinated care is defined by King and Meyer (2006 p.479) as “services that are easy to access and seamless with respect to the interfaces between different services and are tailored to their needs”, whereas service integration is defined as, “functions and activities aimed at the formulation of a unified and comprehensive range of services in a geographical area, where the intent is to enhance the effectiveness of the delivery of services to optimize the use of limited resources”. Recently in Ireland, eight palliative care outreach nurses were appointed to provide an essential role in the coordination and efficacy of care services, acting as the family’s link to other health care professionals and services (O’Brien and Duffy, 2010, DOHC 2010). What is evident from the findings in this study is the important role of hospital nurses, social workers and the Jack & Jill Foundation nurses in connecting parents to services in the community, together with the role of the public health nurse in procuring essential supports for families. Current plans to conduct an evaluation of Ireland’s national policy, for children with palliative care needs is welcomed and should provide further insight into the development of care services (Irish Hospice Foundation, 2014).

7.1.2 The Perpetual Wait for Services

Long waiting times for services forms the fabric of everyday life for families caring for children with LLC's, it can take up to 17 months for needs to be recognized and related services to be arranged. Similar wait times exist in the UK, where parents report waiting up to 18 months to access specialist equipment and occupational therapy (Craft, 2007). Concerns over the supply of equipment was a central issue for parents where they found it difficult to access suitable equipment and experienced extensively long waiting times for services to be provided. These problems are also reported in Townsley et al. (2004) and Steele et al. (2008).

Throughout the duration of 0-4 years, a constant need for the following exists: a key worker, advocacy on behalf of families, rehabilitative services, in particular, physiotherapy, emotional support services, equipment and financial support. Concerns about the future and what will happen after the age of four is also a concerning factor for parents, which became more apparent as their child turned four. This issue is not exclusive to Ireland; a recent study in Scotland, found that families experience delays of between 1-3 years for assessment or services (Stalker et al., 2014) and tightened eligibility criteria, a rise in unmet need and long waiting lists were found to undermine disabled children's rights under international conventions, UK law and Scottish law.

7.1.3 The Crucial Role of the Liaison Nurse

The JJF liaison nurse performs the role of a key worker for families in this study, which includes: providing information, identifying and addressing the needs of all family members, providing emotional and practical support and assistance in dealing with agencies and acting as an advocate; these functions align identically with the description of the key role provided by Cavet (2007). Fundamentally, key workers help to build a package of care for families (Townsley et al., 2004). The need for information, and the issues surrounding it, is a widely reported problem in the literature (Gravelle, 1997, Rahi et al., 2004, Selman et al., 2009, Mitchell and Sloper, 2002). A literature review by Fisher (2001), on the needs of parents with

chronically sick children, identified three main themes: the need for normality and certainty, the need for information, and the need for partnership. The development of a key worker as a central support to families is consistently mentioned as a priority in Ireland (DOHC 2001, DOHC 2005, DOHC 2010, Laura Lynn, 2013) and the UK (DOH 2004, DOH 2005). However, Cavet (2007) points to the fact that there is still no blueprint for the role of the key worker, adding that there is a lack of evidence on the effectiveness of the various key worker models in existence; additionally, they recommend internal and external evaluations in order to further enhance our understanding of this role.

7.1.4 Parents as Advocates

As a result of the gaps and limitations in service provision, parents take on the role of advocates on behalf of their child. Findings from this study reveal that parents invest significant amounts of time conducting administration activities to source, access and organise the provision of care services for their child which is similar to findings from (Nicholl, 2008, Kirk and Glendinning, 2002, Kirk, 1998). The parents in this study developed a thorough “expert” knowledge on their child’s condition, which can be used to educate and inform health care providers. Staying in close contact with key-workers, public health nurses and liaison nurses has also helped to provide parents with helpful information. The gathering of information and problem solving, as demonstrated by parents in this study, has been identified as a positive coping strategy by Beresford (1994).

Mothers, in some cases, contacted the Ombudsman and local politicians to advocate for services on behalf of their child, which may demonstrate an imbalance of power between service providers and parents. Parents in this study felt that if they stopped fighting for services, this would have a direct impact on their child, which is similar to the findings by Wang et al. (2004) on parents’ perceptions of advocacy activities where parents believed it to be an obligation and that the central aim of advocacy was to improve services for their child. Parents also felt they provided a voice for their child that the child could not otherwise provide for themselves, and that advocacy enhanced coping and helped to cause systems change at community and state levels.

Parents described “fighting the system” to be one of the most problematic issues and one that causes more stress than actually caring for the child, which is similar to Resch et al. (2010) where the challenges were not to do with caring, but a result of the absence of environmental supports. Other research by Dybwik et al. (2011) found that parents find the lack of involvement in decision-making and bureaucracy to be a daily struggle. The phenomenon of advocacy, in relation to parents caring for children with LLC’s, constitutes a small but growing literature (Kosik, 1972, Ryan and Cole, 2009, Mlawer, 1993). Advocacy emerged as a central skill, demonstrated by parents, and was developed as a result of a no choice situation; as explained in one mother’s words, “you learn quickly that you have to start shouting and screaming for services”, this too is reported in (Nicholl, 2008, Mlawer, 1993, Kosik, 1972, Dybwik et al., 2011).

Providing information and education to families can improve self-efficacy in managing their child’s care (Longden and Mayer, 2007) and can contribute to lower stress and improved mental health (Soulvie et al., 2012). To further add to this, calls for specialist education training for health care professionals, who are caring for children with life-limiting illnesses, is recognized as a substantial need in Ireland (DOHC 2001, O’Brien and Duffy, 2010, DOHC 2005). As a result of this identified need, two education programmes for health care professionals who are providing care to children with life-limiting conditions have been set up. Both are funded by the Irish Hospice Foundation and managed by Centre of Children’s Nurse Education at Our Lady’s Children’s Hospice, Crumlin (IAPC 2010). However, more investment is needed in improving education curriculums as the lack of postgraduate education is a growing concern in this field (DOHC 2010 p.29).

7.1.5 Physiotherapy – a Very Valued Service

This study identified significant unmet need in relation to physiotherapy services for children with LLC’s. The key concerns reported in relation to physiotherapy include: the lack of service availability which parents felt impedes their child’s development, disparities between the views of parents and health care professionals on their child’s needs and having to privately finance physiotherapy, all of which were similarly reported in the Irish context by Redmond and Richardson (2003).

The disparities between the views of parents and physiotherapists is congruent with the growing body of research, where parents want to have their views and competencies recognized by health care professions (Blackard and Barsh, 1982, Bailey, 1987, Sloper and Turner, 1991, Garshelis and McConnell, 1993, Allen, 2014). Parents also expressed anxiety over having to provide physiotherapy to their child, which they felt further, added to their responsibility and an uncertainty over whether what they were doing helped their child. This is similar to findings from Wiart et al. (2010), on parents' perspectives on physiotherapy, where parents struggled with the demands of providing therapy, along with the daily stresses of caring for their child.

The children in this study typically received 1-2 hours over a four-week period, but more often than not, they received only one hour. Determining the optimal amount to provide is challenging for this patient group, especially when parents call for physiotherapy to be provided intermittently for 10-15 minutes at a time. Findings from a pilot study by Trahan and Malouin (2002) examined the effects of physiotherapy for children with Cerebral Palsy and recommended four treatments per week over a 4-week period, with sufficient rest periods in between. In Ireland's Palliative Care Competence Framework (Ryan, 2014), guidelines for physiotherapists are outlined under the competence domains of: principles of palliative care, communication, optimizing comfort and quality of life, care planning and collaborative practice, loss, grief and bereavement and professional and ethical practice in palliative care. However no specific guidelines are available on suitable provisions of physiotherapy services for children with life-limiting conditions in Ireland.

The descriptive costs study revealed that 52% of the families in this study spend an average combined monthly sum of €95.30 on physiotherapy, occupational therapy and speech and language therapy for their child. This equates to €1,143 annually on rehabilitative services. The remaining 23 families, that reported paying zero costs, may represent a group that cannot afford to personally finance additional services. However, some of the families in this study reported receiving free physiotherapy, after they "ruffled a few feathers". Providing services on this basis is unsatisfactory and demonstrates inequality in the allocation of services.

7.1.6 The Financial Costs of Caring

The descriptive study in phase two provides evidence on the financial impact on families in relation to caring for a child with a LLC. Findings from this study reveal that, on average, parents lose almost half of their post-tax household income (€14,791) due to the additional costs associated with caring for their child. This finding provides evidence of the financial impact on families, where they are essentially impoverished by their circumstances. The findings from this study support the small, albeit growing, literature on out of pocket costs and financial support needs of parents caring for a children with disabilities (Monterosso et al., 2007, Anderson et al., 2007, Revill et al., 2013, Parker et al., 2013). Typical costs of caring include: the purchasing of special equipment, making adaptations to home and car, travel costs for medical appointments and loss of income due to caring for a child full time.

Severity of impairment has been found to increase costs (Martin et al., 1998, Zaidi and Burchardt, 2005, Dobson and Middleton, 1998b) due to the episodic nature of severe conditions, which require frequent hospitalizations, and as a result, higher proportions of income is spent on care. In this study, children have frequent hospitalisations and uncertain care trajectories, however, for the families that reported minimal or zero costs in relation to caring for their child, it is possible that their child's condition is not as severe, resulting in lower out of pocket costs. Geographical location is an important factor to consider and has been highlighted as a key concern in Ireland's policy documents (DOHC 2010, Laura Lynn, 2013). Access to care services, based on geographical location is important when considering the costs associated with traveling to appointments. One family reported having to attend 74 hospital appointments in the previous year. The disparity in service provision in rural and urban areas has cost implications for families, as reported by Smith et al. (2004). These factors, however, were not within the current scope of this study. Nevertheless, they do warrant further research in the future. Although a descriptive study, these results coincide with Dobson and Middleton (1998) work, where they found that it costs triple the amount to raise a disabled child in comparison to a non-disabled child because of the additional direct costs that are incurred, such as adaptations to home, transport, heating, and childcare.

Ireland could potentially benefit from the adoption of a policy whereby parents can use state funding to purchase services on behalf of their child (Redmond and Richardson, 2003). In the UK, and the US, they have adopted such policies, the UK's community care (direct payments) Act 1996 provides families with financial assistance to purchase their own care and support services. Parents can access funding once they have been assessed as needing care services (<http://www.carersuk.org>). Funds can then be used to purchase an extensive range of services, including the purchase of equipment for the disabled person. The aim of the policy is to empower disabled people with the autonomy to purchase services to meet their assessed needs. According to a study conducted in Scotland on the direct payment policy (The Scottish Government, 2013), users with physical disability received funds of up to £15,492 annually. These rates do, however, vary by local authorities in Scotland by their use of direct payments. The direct payment safeguards families against the risk of poverty and provides a greater level of satisfaction and autonomy in relation to meeting their child's needs (Caldwell and Heller, 2003, Heller et al., 1999). However, one must consider recent findings of a review of direct payments in Scotland, by Stalker et al. (2014), where carers appreciated the choice and control it gave but felt local authorities reaped the financial savings, as carers were required to invest time into organisation and administration activities.

7.1.6 Emotional Impact of Caring

Although this study did not specifically examine the emotional impacts of caring for children with LLC's, parents regularly stated that at times, they felt at breaking point and that they needed some form of emotional support which is reported as an area of need by Carnevale et al. (2006) and Steele and Davies (2006). Previous research by Teague et al. (1993) has found that carers can experience high levels of stress, and as a result, family functioning is affected. While there is evidence on the effectiveness of counselling support for bereaved parents, more research is needed on the effectiveness of counselling services to parents along the care trajectory (Knapp and Contro, 2009). This study identified a need for counselling support services for parents, and in particular, found that 51/69 parents expressed a need for

counselling services to help support them cope emotionally, furthermore, 33/69 families were willing to pay €30.00 for a counselling service. In the UK, it cost £1,748 for families caring for with severe life-limiting illnesses to attend four psychologist appointments. The cost of individual appointments ranged from £60.00 - £136.00 (PSSRU 2012). Some of the families in this study reported paying in the region of €30.00 - €60.00 for services from less qualified counsellors, while others reported being unable to financially afford to pay for services. A potential solution identified in this study was for parents to use some of their financial nursing care grant to finance counselling services, however the need for counselling services should be properly addressed and a more robust system put in place to serve these needs. A study by O'Brien (2001) points to evidence on the emotional impact on families who are caring for technology dependent children, where the impact does not become apparent until months or even years later, when the strain of caregiving takes its toll (Diehl et al., 1991, Patterson et al., 1992).

In this present study, referral to emotional support services was demonstrated in some cases by the Jack and Jill foundation nurses. According to Boss and Hutton (2014 p.56), “the medical model for children with LLC’s has developed along traditional diseases specific lines, promoting advances in medical and surgical treatments but the inclusion of developmentally appropriate psychosocial care remains inconsistent”. The free-text comments provided by parents in (Appendix 16) presents further evidence on the need for emotional support services to prevent relationship breakdown which has been found to be a concern for parents in similar situations (O'Brien, 2001). The family system, which is often identified as a potential coping tool, could also be considered as a source of stress; a variety of stressors were identified in one study, including the impact of illness on interfamilial relationships (Soulvie et al., 2012, Hunt et al., 2013a).

7.1.7 A Tentative Future of Care

Concerns over the provision of care for children greater than four years of age, was a universal concern expressed by parents in this study. Parents described the uncertainty around future care provision to be too much to bear at times. They continuously fought to sanction care services in advance of their child turning four

but were consistently met with rejections. This confirms the limited role the state has in the provision of care services in the community (DOHC 2005). In some cases, the JJF continued to provide care and it was evident that the nurses remained in close contact with families even when discharged from receiving JJF grants. The uncertainty around their child's illness, and the additional worry of tentative care plans, makes caring for a child in these circumstances very challenging. Cohen (1993) describes the management of uncertainty by developing strategies to manipulate the known, the unknown and the unknowable which requires parents to manage six interactive dimensions of daily life:

- 1) **Time:** adopting a "one day at a time" philosophy; living in shortened time units, limits uncertainty.
- 2) **Social Interactions:** managing certain aspects of social disclosure can reduce stressful encounters and deciding who to tell, how much and under what circumstances can reduce the profound influences of social interactions that can heighten the awareness of uncertainty.
- 3) **Information:** In some cases, extracting information can be empowering and useful, but in others, knowledge has the potential to incapacitate, thus, relevant strategies should be employed to increase/reduce the amount of information received. In section two, the comments section highlights that parents hear about budget cuts in the media; using this source of information is potentially harmful and may increase unnecessary worry.
- 4) **Awareness:** Pushing the problem to the back of their minds, as stated by parents in this study, or keeping busy and restricting attention to the tasks at hand, allows temporary respite from worry. By managing awareness, parents are able to normalize their lives.
- 5) **Illness:** Constant attention is required by Children with LLC's, so problems can be noticed early and corrective action can be taken. This can be stressful at the beginning for parents, but, eventually, as time progresses, monitoring becomes continual scanning that is focused but built around their developed knowledge of their child's condition. This in turn reduces uncertainty.
- 6) **Environment:** The more tightly that parents are able to control the child's physical environment, the more certain they feel about the child's well-being, with parents being able to make decisions around what is best for their child, even though, not all factors can be controlled.

Further work, by Cohen (1995), highlights triggers that have the potential to bring about heightened levels of uncertainty which includes the routine medical appointments, minor symptoms or variations from the child's norm, specific medical words or phrases, changes in therapeutic regime, evidence of negative outcomes for other children, changes in developmental stages and night-time absence. Although those domains are relevant to families in this study, the key triggers of uncertainty that were more commonly mentioned by parents were in relation to care plans for children after four years, unmet need in relation to rehabilitative services, and financial security.

7.2 PHASE TWO: Preferences of Care Services: Discrete Choice Experiment

Chapter 6 utilized a discrete choice experiment to quantify parent's relative preferences for different service attributes. The results from the Logit analysis revealed that parents have strong preferences for more day nursing care, physiotherapy, speech & language therapy and a cash maintenance payment and are more likely to choose a package of care with those particular services in it.

The strong preference for day nursing care services over night nursing care may be because families use time during the day to conduct other activities in and outside of the family home and also spend time with other family members which they considered to be "normal" family activities, also reported by Carnevale et al. (2008). Findings from the choice experiment also revealed that families with lower income had particular preferences for day nursing care, physiotherapy and a cash maintenance payment, in contrast to higher income families where they were more likely to favour day nursing care, physiotherapy and speech and language. Families living in urban areas had a preference for physiotherapy and both night nursing and day nursing care. This finding may reflect that families located in urban areas live longer distances from other family supports, so these services may serve as a replacement for the lack of informal support. Families living in rural areas had a preference for speech and language therapy, day nursing care and physiotherapy, reflecting perhaps a shortage in some under-resourced rural locations. In Ireland's most recent needs assessment, it was acknowledged that respite care services are

offered to families on the basis of diagnosis and geographic location rather than need (Laura Lynn, 2013 p.43); this indicates that much work is needed to identify projected need for respite services based on age, location and diagnosis.

The mixed logit confirmed the findings discussed above and again the same preferences were found, with night nursing and out of home respite being the least favoured in comparison to the other services. The Latent Class analysis identified two classes of parents with different preferences for care services. The first class of parents would like day nursing care, physiotherapy, speech and language therapy all to form a monthly package of care for their child. Findings from Sloper and Turner (1992) found that parents believed physiotherapy and speech and language therapy to be the greatest areas of unmet need. The second class of parents had a preference for night nursing care and physiotherapy. These findings reveal heterogeneous preferences for caring for a child at home amongst parents, however, class 1 constitutes approximately 80%, which leaves a small percentage of 20% in class 2. The majority of families would like the four services stated above. The huge demand for rehabilitative services for children with LLC's is evident and thus highlights the need for a more active approach by health care professionals to insure families access these services.

The results also indicate some differences in preferences based on socioeconomic characteristics; however, with such a small sample size, caution is necessary. The descriptive costs study provides a backdrop for interpreting the DCE results, and the rationale for parents' preferences. The families in this study bear a large financial burden and preferences revealed in the experiment are not independent of the fact that families have been impoverished by their circumstances. Preferences and choices around care services will be affected by the fact that, almost 50% of families' disposable income is absorbed in the provision of care to their child. The services that are costly and less available like physiotherapy and speech & language therapy are more likely to be preferred by parents, as well as a cash maintenance payment to compensate for the additional costs of caring. Parents are willing to privately finance physiotherapy for their child and in some cases pay up to €500.00 a month, the families that can't afford to privately finance this service fail to receive sufficient amounts for their child. Further analysis on willingness to accept compensation where parents would need to be compensated by the sum of €148.00 for a loss of one hour of physiotherapy and €66.00 for one

hour of speech and language therapy further adds highly that parents value these services. The resources which families have available to them is fundamental to the successful management of their child's care, as families with "high burden and low resources constitute a high risk" (Wong, 1991 p.3).

The findings from the choice experiment revealed that parents would like a slightly different combination of services than they currently receive. An idealized hypothetical monthly care package is presented below in Figure 7.1. This package is not intended to be suited to everyone's circumstances or preferences, but based on the results of this study, presents a monthly care package that represents an improvement over the status quo in most cases.

Figure 7.1 Ideal Monthly Care Package

Night Nurse 24hrs X €16 = €384.00
Day Nurse 60hrs X €16 = €960.00
Cash payment of €150 = €150.00
Physiotherapy 3 X €60 = €180
Speech & Language 3 X €60 = €180
Total Monthly cost per family: €1,854

In a combined monthly package, parents would ideally like the maximum allocation of physiotherapy and cash maintenance payment offered in the choice experiment. They would like to receive 60 hours of day nursing care and 24 hours of night nursing care and the maximum allocation of cash maintenance payment. The estimate for the provisional package would cost €1,854 to provide on a monthly basis and €22,248 annually.

This study is unique in its investigation of the needs of children and their families with life-limiting conditions at home in Ireland. The comprehensive research agenda addressed around service experiences, preferences of care and the financial implications of caring provides many new contributions to the literature. The design of this study facilitated the use of the both qualitative and quantitative

methods, which permitted a more thorough examination of the topics under investigation. Children with life-limiting neurodevelopmental conditions are a distinctive and vulnerable population and require immense input from families on the successful management of care at home. The parents in this study demonstrate enormous strength and resilience and consistently lobby on behalf of their child to procure essential services. The inadequate provision of services, particularly rehabilitative, and the extremely long waiting times for service-set up and equipment provides evidence that current services are not meeting the needs of this population. The results of this study also challenge existing thinking around the need for rehabilitative services in paediatric palliative care. Parent's pay significant out of pocket costs for physiotherapy and the disparity between professional and parent views on supply of this service needs immediate attention. The findings of this study provides compelling evidence on the financial costs associated with caring for a child with a LLC and emphasises the need to urgently address existing financial support mechanisms. Overall, the families in this study call for increased service provision, improved service implementation at the transition from hospital to home phase, more transparency in the allocation of equipment, a link-worker to provide essential information on services, more accessible emotional support services and financial support mechanisms for families caring for children with life-limiting conditions.

7.3 Conclusion

Caring for children with life-limiting neurodevelopmental conditions and their families is a great undertaking that varies in magnitude, intensity and saliency. In phase one of this study the phenomenon of the parents' experience was described, analysed and explicated in order to generate a greater understanding of parents' needs and expectations. Results from this study reveal the nature of caring to be the least stressful aspect of caring, when compared to the battle to procure and maintain services from the moment their child is born right through to when their child turns four. Additional challenges include the extra financial costs associated with caring for their child, reduced employment opportunities and a loss of disposable income. A desired monthly package of care would ideally have the following services,

physiotherapy, speech & language therapy, day nursing care and a monthly cash maintenance payment. In terms of essential supports for parents as carers, counselling was reported as a service that has the potential to ameliorate the stress experienced by parents and help them to manage the emotional impact of caring for their child. The results from this study provides a detailed evidence base for the development of care services that relate to children with life-limiting neurodevelopmental conditions at home in Ireland.

This chapter has provided a discussion of the key findings of this study. The findings relate to the aims and objectives, outlined and presented in Chapter 3. The overall strengths and limitation of the study are presented in Chapter 8.

Chapter 8

Conclusion and Recommendations

8.1 Introduction

The concluding chapter firstly highlights the key messages from the study as outlined in table 8.1. A discussion of the overall strengths and limitations of the study is outlined and the implications and recommendations in relation to Ireland's palliative care policy and the Irish health care system. The chapter concludes with a discussion on the potential future directions of this research.

Table 8.1 Key Messages from This Study

-
- There is a general shortage of services available to families caring for children with LLC's in the community, particularly in relation to rehabilitative services.
 - Parents need more of the following services to meet the needs of their child: Physiotherapy, speech & language therapy, day nursing care and a cash maintenance payment.
 - Parents' experience significantly large out of pocket expenditures in relation to providing care to their child at home, and lose on average up to €14,990 (50%) of their disposable household income.
-

8.2 Overview: Strengths and Limitations of the Study

By specifically focusing on children with life-limiting neurodevelopmental conditions, this study may limit the generalisability to other children with life-limiting conditions; nonetheless, this study represents the needs of an important patient group. The unique approach used to elicit parents preferences may be

usefully applied to other palliative care research. Recruitment bias may have occurred as a result of recruiting through JFJ nurses (gatekeepers) and also the age limit applied, however, in all cases, the families that were excluded were within the strict exclusion criteria, which excluded all families at highly sensitive stages of their child's illness. Furthermore, retrospective accounts allowed the gathering of data on all stages of the child's illness from birth to seven years. In most cases, nurses from the voluntary organization attended interviews alongside the researcher to provide care to the child during the interview process. This may have influenced what mothers were willing to disclose, however, nurses were located in separate rooms to limit potential bias in the parent's accounts.

Some of the mothers in the study had more than one child with a disability, which is not a typical experience of parents in this population group; nonetheless, their experiences were equally as important as other participants and provided a unique insight into experiences. For the interviews, both parents were encouraged to participate, however, only three fathers participated in the interview process, and so, we may have missed some unique perspectives. In all cases however, the primary carer for the child was interviewed.

Some limitations should also be noted in relation to the quantitative study. The results from the statistical models provide estimates of the value and importance to respondents but they do not predict future behaviour or health outcomes (Bridges et al., 2011). The results of this study represent one time point, therefore repeating the experiment at different time points would provide more concrete evidence from which to base recommendations on. A lack of transparency in the qualitative methods, used to define attributes and levels in DCE research, has been reported in other studies (de Bekker-Grob et al., 2012), however, this study employed a rigorous transparent qualitative process to identify and refine service attributes.

Challenges did exist with the population chosen for this study especially in relation to recruitment for the pilot exercises, where many follow up phone calls were made to arrange and re-arrange appointments. One family in particular had to cancel four appointments, all at short notice. This was of course understandable given the nature of their child's condition, however, low participation rates were a constant concern throughout the duration of this study. The use of incentives may

have potentially improved participation rates but in most situations families were unable to attend due to their child's illness, therefore incentives would have been ineffective, insensitive and unethical for this vulnerable population.

During the final data collection (quantitative questionnaire) stage, parents were contacted by telephone to encourage them to return their postal questionnaires. This was found to be an extremely useful exercise, as many parents delighted in sharing feedback with the researcher and subsequently returned their questionnaires promptly. The number of returned questionnaires increased from 30 to 69 questionnaires which was a direct result of connecting with parents by phone. Engaging with parents was a useful way to build rapport in this study and it did result in higher participation rates, which in comparison to other studies in paediatric palliative care, constitutes a very high response rate of 72%.

Finally using a mixed methodological approach provided several advantages, the qualitative data adds to the prevailing knowledge and delivers new viewpoints in health care (Tong et al., 2007) while the quantitative element provides numbers that can be used to add precision to words and narratives (Johnson and Onwuegbuzie, 2004). The descriptive study on the financial costs of caring provided important insight into preferences of care and the rationale for the choices made. Mixed Methods can add insight that may have been missed by using one single method. It produced a more complete knowledge that can be used to inform theory and practice.

8.3 Implications and Recommendations – Introduction

This study identifies key elements of parent's experiences of and preferences of care services on behalf of children with LLC's at home. The findings have implications for a wide-ranging group of strategic planners, service providers, care organizations and policy makers. The implications and recommendations are outlined in the following sections:

8.3.1 Implications for Strategic Planners

- 1) There is a lack of coordination between hospital and community care services, especially with regard to discharge planning and subsequent service set-up (Section 4.6.1).
- 2) There is no evidence of long term care planning for the provision of care services to children with LLC's after four years of age in the community (Section 4.6.8).
- 3) Families face significant financial outlays in relation to caring for their child and the magnitude and impact of this may be underestimated (Section 4.6.6).
- 4) Parents have to make substantial adaptations to cars and houses to accommodate their child's needs, which are costly, unaffordable, and in some cases, non-refundable (Section 4.6.6).
- 5) There is currently no access to the newly formed national database on children with LLC's in Ireland. This makes it difficult to estimate current prevalence rates in Ireland (Section 4.6.6 and Section 6.2.6)
- 6) 8/69 families were not in receipt of a medical card and are potentially experiencing large out of pocket costs as a result (4.6.6).

8.3.2 Recommendations for Strategic Planners

From this study a number of recommendations can be made based upon an analysis of the literature review and the subsequent research carried out within this study:

1. Develop a national plan for the successful management and implementation of transition from hospital to home for families caring for children with LLC's.
2. Develop a national strategy and a specific funding infrastructure for the provision of care services to children after four years of age.

3. Revise and examine existing financial supports for families with the objective of meeting families' needs, especially in relation to the annual respite grant which is too low to meet the current needs of these families.
4. Implement a more efficient system of allocating housing adaptations grants to families who require substantial changes to their cars and homes.
5. Further develop the information system recommended by (DOHC 2010), so prevalence rates can be estimated to aid future care planning and provision.
6. Automatic assignment of medical cards to parents caring for children with a diagnosis of a life-limiting condition.

8.4 Implications for Service Providers

1. The needs of children at the very early stage of diagnosis are not being met and are not clearly identified, with some families waiting up to two years for services to be set up at home (Section 4.6.1 and Section 4.6.2).
2. Equipment is poorly supplied and desperately needed to support children's specific needs at home (Section 4.6.5).
3. There is no evidence of a centralised information point for families (Section 4.6.1).
4. Parents would benefit from emotional support services to help them cope (Section 4.6.1).
5. Physiotherapy services are sparsely provided and difficult to access (Section 4.6.2 and Section 5.16.1).
6. The high cost of special needs toys and equipment needs to be addressed for this population (Section 4.6.6 and Section 6.3.1).

8.4.1 Recommendations for Service Providers

1. Families should have a point of contact in the community upon leaving hospital so they are referred to essential support services and avoid missed opportunities in accessing services.
2. Physiotherapy services need to be provided in a fair and equitable way to all families and guidelines are needed on the optimal amount to provide to these children.
3. One centralised information system is needed to provide information on supports, entitlements, care planning and resources specific to the child's diagnosis.
4. Psychological support services should be offered to parents, not only at the beginning, but also throughout their child's care trajectory.
5. The provision of equipment to families' needs to be allocated in a more timely and equitable way.
6. Subsidisation of special needs toys and equipment is needed and recommendations for costly special needs toys and equipment need to be carefully considered, as families feel under pressure to buy items to support their child.

8.5 Implications for the Voluntary Organization

1. Families are in need of a key-worker at all times (Section 4.6.3).
2. Families would like to use the resources allocated to them by the JJF a little differently, especially to obtain more rehabilitative supports (Section 6.3.1).
3. Families would benefit from more emotional support services (Section 6.5.1).
4. Families that would benefit from using the JJF service are often unaware of the JJF and who the services they provide (Section 4.6.1).

8.5.1 Recommendations for the Voluntary Organization

1. Continue to provide essential key worker roles, communicate this role and the value of it to other health care professionals working with families and create knowledge transfer across other health care organisations and professions.
2. Continue to recommend and source emotional support services for families.
3. Look at ways the existing budget can be used to potentially allocate other resources needed by families.
4. Create more awareness of the JJF and the role they provide; having a referral to the organization at the earlier stages is important.

8.6 Implications for Practitioners Working with Families and Children with LLC's

1. Administration work requires large time investments from parents; this needs to be recognized (Section 4.6.4).
2. Parents feel “isolated” and “cut off” during the transition from hospital to home phase (Section 4.6.1); this too needs to be recognised.
3. Information needs are broad and diverse. Liaison nurses, social workers, general nurses and all other health care professionals need to be informed of what is available for families and / or direct families to one centralised forum (Section 4.6.1).
4. There are misconceptions about the amount of physiotherapy that can and should be provided to children with LLC's and parents are disappointed with poor service provision (Section 4.6.5).
5. All of the valued health care services identified in the DCE experiment need to be reviewed, particularly Physiotherapy and Speech & Language Therapy (Section 5.16).

6. Parents have a high value for physiotherapy and speech and language therapy, which is indicative of unmet need (Section 4.6.4 and Section 7.2).

8.6.1 Recommendations for Practitioners Working with Families and Children with LLC's

1. Support is needed to alleviate parents' administration work load and assistance is needed in negotiating and accessing vital care services.
2. Practitioners need to recognise that families feel isolated when discharged home; communication needs to be maintained and continuous, especially for families who are not yet in receipt of essential services.
3. There is an onus on all health care practitioners to be fully informed on available support services for families to access.
4. A more transparent system of service allocation should be implemented and care plans outlined with families, so they are fully aware of how much of a particular service they will receive.
5. A monthly model of care needs to be flexible and adaptable to meet the varying needs of parents caring for children with LLC's.
6. Guidelines for the provision and allocation of physiotherapy and speech and language therapy needs to be developed.

8.7 Recommendations for Policy Makers

1. Families caring for a child with life-limiting neurodevelopmental conditions should have automatic assignment of a medical card.
2. The annual respite grant reductions need to be revised, particularly in relation to 2012's 20% budget reduction.
3. Continuous needs assessments are necessary to address the significant gaps in service provision for children with LLC's.

4. The provision of care services for children greater than four years of age needs immediate attention.

8.8 Recommendations for Future Research

The findings of the current study have highlighted a number of potential areas that could be addressed by future research. Examining father's experiences, and particularly the differences between both mothers and fathers, would help to further explain the experiences of caring for a child with a LLC. In relation to services, the parents in this study had mixed views on out of home respite care, some were indifferent, while others used the service frequently. It is possible that geographic proximity plays a role in accessing out of home respite care so it would be useful to explore this further in the Irish context.

A key issue for families in this study is the cessation of care services after their child turns four years of age. There was no evidence of care plans for the future and arbitrary age limits on entitlements to care needs to be abolished for this population. Developing a national plan for the provision of care services for children with LLC's which encompasses all age groups would be a useful extension of this work, especially as this study has provided useful evidence on what parents want and need more of. There is also a need to conduct more research to bridge the gap between parent's expectations and those of service providers particularly in relation to rehabilitative services.

Identifying effective ways to communicate to parents through technology is a useful way of identifying unmet needs at the very early stages of diagnosis and throughout the child's illness trajectory. There is a need not only for more evidence on the costs of caring for children with LLC's but also for the development of strict criteria to measure costs. Finally, it is clear from the findings of this study that families have different needs at different stages, it would be useful to conduct this study with a larger sample to define the specific needs based on the child's age and severity of diagnosis.

8.9 Conclusion

The current study provides new insight into the experiences of parent's caring for children with life-limiting neurodevelopmental conditions at home in Ireland. The mixed methods approach provided a comprehensive and reliable evidence base on which to inform current service planning and provision. The findings from this study presents evidence on the needs of families caring for children with LLC's at home. Key areas of need include: rehabilitative services, especially physiotherapy and speech & language therapy and financial support to help with the additional costs of caring. Parents would ideally like a package of care that is flexible and includes more of the following services; physiotherapy, speech & language therapy, day nursing care and a cash maintenance payment on a monthly basis. A key finding is that parents are willing to forgo some services in order to receive more of others. Parents would also value more emotional support in the form of counselling and there is a need a centralised information system for families to access information about services and entitlements. In the current economic climate, where resources are very limited and budget reductions prevail, it is now more important than ever to use existing resources more efficiently. This study makes many effective recommendations that in some cases require little to no financial outlays. The more costly recommendations however, remain challenging, especially with current reductions to the health care budgets. Ireland's Palliative care policy agenda has many nuances and as a model is still in early development, particularly in relation to children's palliative care. These findings are timely and can be used to inform current policy especially, as this area is now receiving more attention.

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Appendices

Appendix 1 - Overview of the HRB SPHeRe PhD

Overview of the HRB Structured Population and Health Services Research Education (SPHeRE) PhD Programme

Programme structure

Scholars are provided with a structured PhD training programme, consisting of 4 training strands with strand 1 completed in Year 1 and strands 2-4 extending over 4 years. The training in strand one is equivalent to a Masters level programme in health services research. The training in strands 2-4 addresses specialist research skills related to the scholar's research project, professional development and career development skills. A particular feature of the planned training is the determination to ensure that graduates of the programme are part of a network of HSR specialists that will provide mutual support and facilitate collaborations over their careers.

The four formal training strands are:

1. One year intensive course in Health Services Research
2. Specialist research skills training (related to research project)
3. Professional skills including time and project management, grant application skills, scientific paper writing, team-working, communication skills (written and oral, including media and advocacy training) and presentation skills
4. Career development skills (CV and interview skills, seminars with experts and leaders in relevant career settings)

Overview of year one:

Year 1 of the programme includes classroom, online and self-directed learning, workshops and seminars and the completion of a national placement in a research organization. The aim of the national placement is to provide understanding of how research is carried out, how data is collected and accessed, and how research is disseminated and put into policy and practice. A significant element of self-directed e learning is required and the programme avails of RCSI's e-learning infrastructure. By the end of Year 1, students develop a draft PhD proposal (including literature review, methods and feasibility issues addressed).

Core Modules:

Modules are listed below covering contextual knowledge (particularly on health systems and

population health), information systems and sources, generic research skills such as reviewing evidence, introductory material covering the main HSR disciplines and courses in discipline-based research.

Module 1: Population and Individual Health

Module 2: Health Systems Policies and Health Informatics

Module 3: Evidence Synthesis and Clinical Trials

Module 4: Applying Research Methods

Module 5: Data Analysis with STATA

Module 6: Health Economics and Econometrics

Years 2 – 4

There is ongoing evaluation, peer support and course seminars for scholars throughout years 2-4. At the end of year 2 and year 3 the scholar and the supervisor submit evaluation reports. These are assessed with reference to the PhD proposal and the Steering Sub-Group gives feedback to the PhD scholar and the supervisor.

Appendix 2 - Ethics Approval Letter



COLÁISTE NA TRÍONÓIDE, BAILE ÁTHA CLIATH | TRINITY COLLEGE DUBLIN
Ollscoil Átha Cliath | The University of Dublin

Ms. Aoife McNamara,
Department of Health Policy and Management,
3-4, Foster Place,
Trinity College,
Dublin 2.

28th January 2012

Re: "Preferences of care for children with life-limiting disabilities in Ireland" A Discrete Choice Experiment

Application 12/010/2011

Dear Aoife,

Thank you for your submission of the above proposal to the HPM/CGH REC.

The REC has given ethical approval to the proposed study.

Prof. Charles Normand
Chair of the HPM/CGH REC

An Coiste um Thaighde Eitice
*An Lárionad Pholasaí agus Bhainistíocht
Sláinte/Lárionad na Sláinte Domhanda*
3-4 Plás Foster, Seomra 0.18,
Coláiste na Tríonóide,
Baile Átha Cliath 2, Éire

Research Ethics Committee
*Centre for Health Policy and
Management/Centre for Global Health*
3-4 Foster Place, Room 0.18,
Trinity College,
Dublin 2, Ireland

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hsmsec@tcd.ie
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health_policy_management/

Appendix 3 – JJF Ethics Approval letter



*Jack & Jill went up the hill,
To fetch a pail of water,
Jack fell down,
& broke his crown,
& Jill came tumbling after.*

*Then up Jack got & home did trot,
As fast as he could caper,
To old Dame Boggan,
Who patched his noggin,
With vinegar & brown paper.*



1st February 2012

Ms Aoife McNamara
Department of Health Policy and Management
3-4 Foster Place
Trinity College
Dublin 2

Re: "Preferences of care for children with life-limiting disabilities in Ireland" A Discrete Choice Experiment"

Dear Aoife,

Thank you for your application for ethical approval from the Jack & Jill Children's Foundation for the above research.

We are delighted to give ethical approval for the proposed study.

Yours sincerely,

A handwritten signature in black ink, appearing to read 'Jonathan Irwin'. Below the signature is a horizontal line and the text 'Jonathan Irwin C.E.O.'.

CEO: Jonathan Irwin FPPAED (1668). Chairman: David Reid Scott. Patron: Eiddie Hobbs. Company Secretary: Patrick Dawson.
Board Members: Dan Murphy, Dolores Molloy, John Sheridan, Donal Kavanagh, Senator Mary Ann O'Brien, Dr. Fin Bresnahan.
Friends of Jack & Jill: Tim Hynes, Mick Kavanagh, Charlie Marless, Peter Murray, Aedin O'Leary, Oliver Sutherland, Jo Symons, Bernard Walsh.
THE JACK & JILL FOUNDATION is a registered Charity - CHY No. 12405.
Johnstown Manor, Johnstown, Naas, Co. Kildare, Ireland. Tel: +353 45 894538 Fax: +353 45 894558.
Email: info@jackandjill.ie Website: www.jackandjill.ie

Appendix 4 - Letter of Invitation from voluntary organization to families

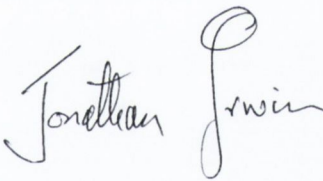
23rd June 2013

Dear Parent/Parents

I wrote to you last year asking for you to participate in an in-depth study into the Jack and Jill service called “Preference of care for children with complex needs”. I am very grateful to you for agreeing to participate in this study. The questionnaire is now ready to be sent out to you and I would be very grateful if you could fill out the questionnaire and return in the envelope provided within ten days.

The study is very important to the Jack and Jill Children’s Foundation, as it will increase the awareness about Jack and Jill and the service we offer. It will also help us lobby the government on your behalf on the preference of care packages you wish to have for your child. Thank you for your assistance on this very important piece of research.

Best regards,

A handwritten signature in cursive script that reads "Jonathan Irwin". The signature is written in black ink and is positioned above a horizontal line.

Jonathan Irwin
C.E.O.

Appendix 5 - Recruitment: Letter of Invitation to Potential Participants

1 May 2012

Dear Parent/Parents,

We are conducting research that explores parents' preferences of care for children with complex care needs. We hope to gather knowledge about the services that matter most to parents and children in these challenging circumstances and identify packages of care that are most suitable. We hope by obtaining parents feedback on preferences of care, we will help researchers and service providers further understand patient needs better. It will also provide valuable information on the best way to organize resources in a more satisfactory and efficient way.

If you are a parent of a child receiving care from the Jack & Jill foundation and wish to participate please read the **Participant Information Leaflet** attached. This will provide more detailed information on the research project and what will be required of you should you wish to take part.

Important note: If you are willing to take part in this study, you will need to sign the consent form attached and return the form in the stamped addressed envelope provided, no later than May 11th 2012.

This research is being undertaken by Aoife McNamara and Professor Charles Normand at the Department of Health Policy & Management, Trinity College Dublin.

Thank you for taking the time to read this

Best regards,

Aoife McNamara

Appendix 6 - Recruitment: Participant Information Leaflet

Title of study:

“Exploring Service Use and Preferences of Care for Children with Complex Care Needs and their Families in Ireland – A Mixed Methods Study.”

Name of Investigator: Aoife McNamara

Name of Supervisors: Professor Charles Normand & Dr. Honor Nicholl

This study is conducted in partial fulfillment of the Health Research Board funded PhD in Health Services Research.

Introduction:

Participants are being recruited to take part in a research study exploring preferences of care for children with complex care needs who are receiving care at home in the republic of Ireland. Before you decide if you wish to take part, it is important that you understand why the research is being conducted and what will be involved. Please take time to read the following information with care, and if you wish, feel free to discuss with friends, relatives and the researcher. If there is anything that seems unclear, or you would like to know more about, please ask. Take time to consider if you do or do not wish to participate. Thank you for taking the time to read this document.

What is the purpose of this study?

The study aims to develop further understanding about parents’ preferences of care for children with complex care needs. We hope to find out what services matter most to parents and children in these

challenging circumstances and identify packages of care that are most suitable. We hope by obtaining parents feedback on preferences of care, we will help researchers and service providers further understand patient needs better. It will also provide valuable information on the best way to organize resources in a more satisfactory and efficient way.

What will happen if I take part/ what do I have to do?

If you agree to take part: a random selection of participants will be asked to meet with the researcher (at a place of convenience for them) to take part in a semi-structured interview. This researcher will discuss a number of topics with you including; how the services are provided, the type of services you receive, concerns you may have had with aspects of service provision and also a general discussion around care needs. The aim of the semi-structured interview is to gather as much information as possible about the typical weekly package of care your child

receives. The researcher will then use the information gathered from the semi-structured interview to design a more formal questionnaire.

The formal questionnaire is a particular type of questionnaire used in health services research. We plan to administer the questionnaire to all of the Jack and Jill parents who agree to participate in the study. The formal questionnaire will present you with hypothetically designed packages of care (i.e., a typical weekly package of care) and you then must decide between each alternative package of care choosing the one you most find suits the needs of your child and family. Instructions will be provided on how to fill out the questionnaire and the researcher will be happy to call you to go through the questionnaire and answer any questions you may have. This questionnaire will be sent to all families by post with a stamped addressed envelope for return.

In order to make sure the formal questionnaire is of optimal quality, we will pilot some of them before the final administration outlined above. This will allow us to test the understanding of the questionnaire from the participants' perspective. We again will select a random sample of participants and send them a pilot questionnaire with a stamped addressed envelope for return. We will then check the validity of the questionnaire and make changes where necessary before the final administration.

Where will the semi-structured interview take place?

The researcher will discuss with you a suitable venue for the semi-structured interview. This will be made as accessible as possible. If you incur any traveling expenses, (these will be refunded within reason). The researcher has requested to accompany the Jack & Jill foundation nurses to family homes to conduct these interviews as this would be more convenient for the participants. A consent form is attached for you to agree to have the interview conducted at your home.

What are the possible benefits of taking part?

In participating in this study, you will have an opportunity to talk about your preferences of care for your child. You will be helping to advance knowledge and understanding of the organization and delivery of care services at home for your child and others. The care provided by the Jack & Jill foundation is the only home care model in Ireland and this will give us an opportunity to gain understanding into what works best and what does not. You will also contribute to a limited body of research on patient preferences of care in relation to children with complex care needs.

What are the possible disadvantages and risks in taking part?

The disadvantages and risks involved are minimal. The Questionnaire may take a half an hour to complete so therefore it may be a little time consuming for already busy parents.

Should I take part?

It is your choice to decide whether you wish or wish not to take part. If you decide to take part, you will be given this information sheet to keep and you will be asked to sign a consent form, a signed copy of which you will receive. You are free to withdraw at any time without giving any reason.

Will my taking part in this study be kept confidential?

Your identity will remain confidential. Your name will not be published and will not be disclosed to anyone outside the study group. Instead of your name, you have a unique research code number. All of the information will be anonymised and held in a secure office locked filing cabinet.

What will happen to the results of the research study?

It is anticipated that the results of this study will be published in a scientific journal and the researcher would be happy to inform you of where to obtain a copy of the published results when this happens. The result may also be potentially represented at Health Services Research conferences. The results will be used to advocate on behalf of the Jack and Jill foundation for an optimal package of care. In all instances, you will remain anonymous and all the information remains confidential.

Voluntary participation:

If you decide to volunteer to participate in this study, you may withdraw at any time, if you decide not to participate.

Who is organizing the research?

My name is Aoife McNamara and I am a postgraduate student at the Department of Health policy & Management, Trinity College Dublin. I am doing this study as part of my work for my PhD. My motivation for doing this study comes from my interest in the organization of and delivery of care for children with complex care needs. I am also interested in gathering patient views on care in order to potentially incorporate their feedback into service planning and provision. The aim of this study is to obtain parents' feedback on particular aspects of care that

matter most to them when it comes to care for their child. We also want to give parents a voice and a role in the way services are organized and delivered.

Who has reviewed this study?

The Department of Health Policy and Management ethics committee have reviewed this study. Professor Charles Normand will be reviewing this study on a regular basis throughout my PhD.

Permission: Ethics approval has been gained from the Research Ethics Committee, Trinity College Dublin.

Contact for further information:

For further information about this research study, please feel free to contact Aoife McNamara on 087 7858937 (9.30am – 5pm). Alternatively you can contact mcnamaao@tcd.ie

Appendix 7 - Recruitment: Consent Form

CONSENT FORM: To be signed and returned in stamped addressed envelope attached.

CONSENT FORM FOR CONTRIBUTOR NUMBER:

PROJECT TITLE:

“Exploring Service Use and Preferences of Care for Children with Complex Care Needs and their Families in Ireland – A Mixed Methods Study.”

PRINCIPLE INVESTIGATORS: Principle Investigators: Aoife McNamara and Professor Charles Normand, School of Medicine, department of Health Policy & Management, Trinity College Dublin.

BACKGROUND: The aim of this study is to elicit preferences of care from parents of children with complex care needs receiving care at home through the Jack and Jill Children’s foundation. The study is composed of two parts; the first part requires you to complete a semi-structured interview in your home or a place that is convenient to you. The interview will last 30 – 45 minutes in duration. A random selection of participants will be asked to take part in this interview so not all parents will be contacted at this stage. The interviews will be used to gather information about your experiences and preferences of care for your child. The second part of the study requires you to fill out a postal questionnaire which will gather more information about your preferences of care and identify packages of care most suitable to your child’s care needs. The questionnaire should take no longer than 30 minutes to complete and will be sent to all parents willing to participate in this study.

In order to protect the integrity of the research process all interviews will be dealt with in the following way:

- The interview will be digitally recorded on a laptop and also audio-recorded using a cassette device. Recording the interview is necessary so that I can accurately reflect on our discussion. The recordings will only be reviewed by myself and my research supervisor Professor Charles Normand; such that they may be transcribed and analysed. The recordings will then be destroyed.
- Upon request contributors can be provided with a full transcript of the interview and are free to change or withdraw any information shared in the interview.

- All recordings and subsequent transcripts will be anonymously labelled and will not be used for any purpose other than that of this study. The data will be confidential and stored in a secure manner in keeping with the requirements of the Ethics Committee of Trinity College.
- All views shared by contributors will be treated confidentially and all comments will be reported anonymously. The information gathered during the study will not be used in future unrelated studies without your specific permission.

DECLARATION:

I have read, or had read to me, the information leaflet for this project and I understand the contents. I have had the opportunity to ask questions and all my questions have been answered to my satisfaction. I freely and voluntarily agree to be part of this research study, though without prejudice to my legal and ethical rights. I understand that I may withdraw from the study at any time and I have received a copy of this agreement.

Childs name [Please print name in block letters]:

Parents name [Please print name in block letters]:

Contact details:

Participant's signature:

Date:

Statement of investigator's responsibility: I have explained the nature and purpose of this research study, the procedures to be undertaken and any risks that may be involved. I have offered to answer any questions and fully answered such questions. I believe that the participant understands my explanation and has freely given informed consent.

INVESTIGATOR'S SIGNATURE:..... Date

Appendix 8 - Letter of Application to Use Premises

Aoife McNamara
Dept. Health policy &
Management,
3-4 Foster Place
Trinity College
Dublin 2
Tel (01) 8962762
16TH March 2012

The Jack & Jill foundation
Johnstown Manor
Johnstown
Naas
Co Kildare

RE: APPLICATION FOR USE OF PARTICIPANTS HOME.

Dear Participant,

Further to my application to complete a research study with the Jack & Jill Foundation, I wish to seek approval to visit you at your home or a place convenient for you to complete a semi-structured interview.

As outlined in the research proposal a location convenient for the participant is required if participants opt to complete the interview. The participant's home may be a convenient location as they are understandably under time pressure. You will receive notice in advance of the dates of interviews should you agree to participate. Please sign the consent form attached. Thank you for considering this request

Yours sincerely,

Aoife McNamara

Appendix 9 - IPA Interview Schedule

Model of Care at Home for Children with Life-limiting Conditions

Interview Theme sheet

Unique identifier number:

- Respondent has read the information sheet.
- The information sheet was discussed with the respondent.
- Confidentiality and data handling procedures have been discussed.
- Consent sheet has been signed before the interview.
- Permission for recording the interview has been given

Theme 1: Demographics and Background

Age / Sex of child/ D.O.B:

Nature of illness / Diagnosis:

Size of family (more than one child with a disability?):

Employment status: how has the situation changed?

Geographic location:

1. Tell me about your child? What age were they when they were first diagnosed?
2. How long did your child spend in hospital?
3. What advice did you receive about the services available to you?
4. Who provided this information?

Theme 2: Pathways to care / Description of services

Jack & Jill Foundation

Now I would like to discuss the Jack & Jill Foundation...

1. How did you hear about the Jack & Jill foundation? Who informed you?
2. At what stage of your child’s illness did you hear of the (JJF)?
3. When you found the JJF, what happened then?
4. How did you begin receiving services from them?
5. What age was your child when they started receiving care from the JJF?
6. What does the service provide?
7. What is your allocated budget?
8. Has this budget changed since the recession?
9. What services do you pay for with your budget?
10. How many hours of each service do you receive weekly/monthly?

JJF Model:

Service Provider / Funding Body	Service Type	Actual Hours Weekly	Actual Hours Monthly	Desired hours weekly/monthly
JJF	Nurse			
JJF	Carer			

11. How long is your child receiving care from the foundation?

12. What other services do the JJF provide to you?

Other Services...

1. Before obtaining care from the Jack & Jill Foundation? What other services did you receive?
2. How was that different to the Jack & Jill Foundation package of care?
3. How often would your child visit hospital? On average how long would your child stay on each visit? What is the longest amount of time your child has spent in hospital?
4. Do you receive any other form of help such as; PCCC from the HSE or care / support from any other voluntary organisation?

Service Provider	Service type	Actual Hours Weekly	Actual Hours Monthly	Desired Hours Weekly/ Monthly
HSE?	Nurse			
EI / BOI?	Carer			
	Physiotherapy			
	Occupational therapy			
	Speech & language			
	Social worker			
	Disability Liaison			
	Public health nurse			
	GP			
	Counselling services			
	Play therapist			
	Home teacher			
	Dentist			
	Orthotics			
	Special needs assistant			
	Paediatric outreach nurse			
	Emergency services			
	Public health nurse			
	Community welfare officer			

Laura Lynn	Out of Home Respite			
---------------	---------------------	--	--	--

5. Do you receive any other financial support from the following?

- Charity organisation?
- Government payment/ grant?
- Family?

6. Who are the other main sources of support? Family / Friends etc.? In what way do they support?

7. Who are the primary carers for your child? Parents / Nurses etc.?

8. If your child is sick, who is your first point of contact?

- a. GP:
- b. JF nurse:
- c. A&E:

Theme 3: Satisfaction with services:

1. How would you currently rate the overall service your child receives:

- a. *Excellent,*
- b. *Good*
- c. *Adequate*
- d. *Poor?*

Can you tell me why you rated the service that way?

2. Apart from the services you get, is there anything else that you think your child needs, that you do not currently receive?

3. Are there any services you feel could be availed of instead of another?

4. In your opinion, what are the top three services most important to you and your child?

5. Do you find any of the services provided unsatisfactory? i.e., not enough of the service provided. What do you think can be done to improve it?

6. Is there any way you would change the current service provided? i.e., package of care received / individual services provided?

7. Your child is years of age, have you any concerns about future provision of care services?

8. What are your main concerns about the provision of care for your child for now / or in the future?
 - a. *in the past?*
 - b. *at this current stage?*
 - c. *for the future*

Theme 4: Family Experiences and Coping

1. What do you do to help you deal with the stress of looking after your child?
2. What support would you personally like to help you cope? E.g., counselling, time to yourself. Activities outside the care setting.
3. Do you have contact with other parents in similar circumstances? If so, do you find this helpful?
4. Can you describe what is the most challenging aspect of looking after your child?
5. How do you feel your child's illness has impacted family life?
6. Have you or your partner had to give up work to look after your child? If so, what has changed? How many hours less/more?
7. Are you finding it difficult to manage financially? If so, what extra costs are directly incurred by you in providing care for your child? And where do these costs come from? E.g, Technology for your child.
8. Finally are there any other concerns or issues you would like to discuss further today?
9. Are you happy with everything you have disclosed today?

CONSENT FORM HAS BEEN SIGNED [TICK BOX]

Appendix 10 - Reflexive Journal

Reflexive Journal:

“Expert carer”

Parents become “expert” carers for their children and articulate the complexities of their child’s illness like a medical professional. The nurses explained that parents become more confident with time, as they gather more and more information and often become the best candidates to decide on what their child needs. Some parents are more confident and outspoken than others. At the beginning of their journey at initial diagnosis, parents describe the initial shock they experienced and wanting to bring their child home. As time goes on, they learn more and develop terminology around their child's condition; this in turn enhances their decision-making ability and autonomy in the case of looking after their sick child. One parent described how they are fully involved in the care plan for their child and how they consult confidently with the nurses and doctors. Other parents describe less positive experiences and often said they did not feel “in control” but that it gets better with time as they become more experienced.

“Some more willing to except help than others”

Some parents are more open to receiving forms of support (carer, nurse, finance, psychological support, contact with other parents) than others. There is a clear difference between the parents that accept help from those that do not. The parents that accept help seem more confident and satisfied, whereas those that do not, seem stretched and have a lack of trust in anyone else but themselves to care for their child. As a result of this, they seem to struggle to cope much more.

“Impact on family”

Some of the parents said they felt guilty about the other children, as all of the decisions were made around their sick child. One mother described using respite as a day where she takes her healthy children out to do something like a normal family would. Some parents said that other children in the family are required to have a lot of understanding that would not be normally expected at their age. One mother used the term “cooped up”, where her child had to regularly stay at home for long periods of time because the sick child was unable to leave the house. Some siblings were more accepting of their sick sibling than others, where they take on a protective role or do not want to be involved at all. One parent described the first year with her sick child as very difficult on her other children. She said she thought they were doing fine but their grades started to really suffer in school. When she talked to them, she realised they were

struggling. She said all she could think of first was looking after her sick child and to get him home for good. It was difficult for the mother trying to manage all of her family's needs in the midst of a major crisis, with the impact on the family being only observed when things calmed down.

“Relationship breakdown”

Some parents expressed a concern that their relationship struggled because of the challenges they faced in caring for a sick child. One parent described getting up every 20 minutes with their sick child the sleep deprivation that ensued. Her husband was working nights (to support the family financially) and therefore slept during the day. This often left them with little time together which resulted in the mother feeling very isolated. One mother explained that her husband was so stressed she told him to ask a GP about getting some psychological help. The GP referred him to a local place that dealt with alcohol addictions; subsequently, her husband never followed up nor did the GP. She felt this was a very crucial stage for them and that one of them needed help. She felt let down by the system as a result.

“Location”

Geographic location was very important and really made a difference when accessing services. Rural areas have less community services resulting in large disparities in service provision. Families have often moved to a different location or expressed a need to be in a more suitable location. Some feel locked in however, due to paying a mortgage in their current location.

“Equipment”

Equipment is a major issue for these families and is raised again and again by parents. There is poor access and long waiting times to receive equipment and in some cases when it does arrive the child has outgrown it.

“Sleep deprivation”

Parents often describe getting little to no sleep and say this can cause problems/tensions at home.

“Care after four years of age”

When asked about the things that worry them most about the future, parents express a worry about continuity of care. Some of the parents said that they don't think about the future and take each day as it comes. Others talk about the battles they have with the HSE to procure care for their child.

“Louder you shout.... the more you get”

Parents say they invest significant amounts of time calling care providers trying to procure services for their child. Only in a few cases, did parents mention investing little to no time and these were the less severe cases. It seems to me that those who shout the loudest about what they need are quite often the families with more support services for their child. Some parents talked about how happy they were with what they have and felt very grateful. One parent described feeling guilty about getting support, as she knew she was taking that support from another family. Some of the parents with stronger personalities said they regularly argued with HSE staff to obtain the care services they felt their child deserved. What is concerning from my view point is that those parents who are shy about asking for things seem to receive less, however, the liaison nurse does remain in contact with families and observes closely what families need which mitigates this concern. The liaison nurses were described as “life savers” on many occasions as they often negotiate on behalf of parents. This was seen as a brilliant support and often took the pressure off the families.

“Poverty”

Some of the parents described at the start of the interview that they were wondering how they would go about defaulting their mortgage. They had purchased their house before their child was born and it no longer suited the needs of their child. In some cases, parents are carrying their child upstairs to sleep at night and because their child is growing all of the time this places a significant burden on their health. They said their house is simply too small and how they needed a downstairs bedroom for their sick child. Most parents expressed a need for financial support in relation to adapting their home to meet their child's needs.

“Support offered to some families and not others”

One woman described being in the hospital with their child and described getting “one blow after another” from the Doctor about their child's health. No services were offered to her and she described how one woman who had a less sick child was having more of a fuss made of her. She felt that because her child was so sick and it looked like there was no hope, no one bothered to offer them anything, not even counselling.

“Family support”

Family support in the community (or close proximity) is very important and some parents say they wouldn't survive without it. There was a 50/50 divide amongst families, some expressed continuous support by parents and brothers and sisters and others expressed no support at all. The reasons for no support include; family members were nervous around the sick child, they were afraid, they didn't feel confident being left alone with the child. Location was often raised in relation to obtaining family support. One family talked about being in negative equity and had no choice but to stay where they were and that if given the choice, they would move closer to family as a support.

“My own experience as a researcher”

Interviewing families caring for children who were suffering from life-limiting conditions was an extremely challenging but highly rewarding experience for me. The strength and resilience demonstrated by the families and the positivity they expressed in the face of adversity inspired me immensely. Throughout the interviews and the transcription phase, I was deeply saddened by some of things that parents disclosed; in particular the initial shock of receiving the diagnosis, the issues around accessing care, and then enormity of parents' roles in providing caring to their child. The parents in this study experienced enormous life-changing events and some of the ways they expressed it will stay with me forever. I found it useful to space out the interviews and conduct them slowly over the period of 12 months so I could better cope with what families had disclosed. In all situations families demonstrated the greatest love and devotion for their child and it made me appreciate with greater clarity what constitutes a real challenge in life.

Appendix 11 - Sample of IPA Analysis

Interview Excerpt	Themes Identified
<u>Equipment set-up & Timing:</u>	
“I was a year waiting on a buggy, which is ridiculous. I am now two years waiting on a chair”	<ul style="list-style-type: none">• <i>Timing issues with getting access to equipment</i>
“The seating is another story altogether. He’s in that chair for three years and we’re still fighting to get him a new one.... we can’t bathe him. There’s nothing there, it’s not even the funding. There is no equipment suitable to bring in to a normal house to bathe a child like mine. Anything you put in the bath is going to hurt him. I mean we just take him on our lap and bathe him ourselves.”	<ul style="list-style-type: none">• <i>Long length of time in wrong equipment</i>• <i>Fighting for services</i>• <i>Correct equipment not available</i>• <i>Concerns over existing circumstances – regular bath could potentially hurt child</i>• <i>No choice but to bathe child on lap themselves</i>
<u>The Fight For Services:</u>	
“Chasing services, it’s so frustrating. It’s not clear-cut. They should make it clear what’s there and if there are people that have more needs, then they should be looked after first. They should make it a priority-based system if resources are tight. It shouldn’t be who screams the loudest. It’s just not fair.”	<ul style="list-style-type: none">• <i>Chasing services</i>• <i>No systematic way of finding service supports</i>• <i>Priority based system needed</i>• <i>A fairer system needed to appraise and allocate services to families in need</i>
“I won’t stop until I am getting the	

services, I am fighting for them. I set out an hour every day to make the calls. Some parents get so tired and I do, because you feel that you shouldn't have to be fighting for this service. Then I think it's best to keep going because what will happen to her if I let it get to me."

"If you don't ask, you won't get with (name of organization), I had to ruffle a few feathers and get them annoyed and next thing I get this number for this service and it's free."

"Not having to fight the system that's geared to say no and not dealing with six different people to get a buggy. The budgets mean they are always pushing you forward to the next budget year."

"There's so much to organise! We went from worrying about changing nappies to managing a tracheostomy child. The part that's gruelling is not looking after our child, it dealing with the health care system."

Physiotherapy – a highly valued service:

"From what I understood, I thought Physiotherapy was going to be a big part

- *Prioritizing time to negotiate access to services*
- *Fighting for services*
- *Fatigue in the push for services*
- *Anxiety about what will happen if she gives up the battling for services*

- *Obtaining services through ruffling feathers i.e., (kicking up a fuss).*
- *A battle for services*

- *Us against them i.e., families vs. the State*
- *Frustration of dealing with myriad of people to obtain a service*
- *Being fobbed off because of budgets*

- *Challenges not with looking after child but dealing with the health care system itself.*

- *Experience of services provision different from what was expected*
- *Frustration at how unfair it is having*

of my daughter's life and that we would be in and out to (organization name) once or twice a week. Like they expect me to learn it and do it with her but that's not fair, I'm not a Physiotherapist, I don't know if what I'm doing helps her."

"The whole Physiotherapy thing is wrong, I feel like no parent should have to do the Physiotherapy themselves, I mean if that child is not developing properly, then that's my fault. No one is keeping tabs on the progress she makes from it."

"I will be spending some money on private physiotherapy."

"She doesn't get as much to reach her full potential, her hamstrings are tight and they need to be worked on every day. She gets tired, so I have to do it for 2-3 minutes and give her a break, I mean you can't just do a one hour physio session... it's not like that. I need more of that kind of help, more frequency is what I really need."

to do the job of a physiotherapist

- *Uncertainty if what she is doing helps child*
 - *Parent's frustration at having to do physiotherapy themselves*

 - *Responsibility for child's physical progression an added stress*
 - *Nobody cares to follow up*

 - *Parents privately finance physiotherapy because of poor service provision*

 - *Physiotherapy enables the child to reach full potential*
 - *Physiotherapy needs different for children with disabilities - only small amount required.*
 - *More frequency of Physiotherapy service needed*
-

Appendix 13 - Discrete Choice Experiment Questionnaire

Questionnaire about parents' preferences of care for children with Complex care needs at home in Ireland. (POC STUDY)

Dear parent,

The aim of this questionnaire is to gather data on parents' preferences of care services for children with complex care needs at home. We are also interested in your current situation such as the additional financial expenses you face and additional support services you may need. You are receiving this questionnaire because you have signed a consent form to participate in this study. Participation in this study is entirely voluntary. The results will be treated as strictly confidential and will be used for research purposes only. Phase one of this study is already complete. I have met with 25 families (randomly selected) to gather data on their preferences of care and, using this data, I developed the final questionnaire attached.

There are two sections in this questionnaire to complete. We realise it might take some time (approx. 10-15 minutes) to complete but this research is vitally important for the development of service provision for families, the Jack & Jill Foundation and other care providers in the future.

Detailed Instructions on how to complete this questionnaire are on page 2.

Please feel free to contact me with any questions at any time on 086 0636257 or please contact your JIF Liaison nurse. Thank you.

This study is supported by the Jack & Jill Foundation.



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COLÁISTE NA TRÍONÓIDE, BAILE ÁTHA CLIATH

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UNIVERSITY
OF DUBLIN



INSTRUCTIONS FOR SECTION 1 & 2:

Section 1:

Presents you with 12 different choice sets. In each of the 12 choice sets, there are two different monthly care packages, package A and package B. Each monthly care package offers six different services. The amount of each service offered varies in each package. You are required to compare package A and package B and select the one you **prefer most** in each of the 12 choice sets. The questionnaire is clearly labelled to help you work through each choice set.

Description of care services in each package:

Hours of night nursing care provided on a monthly basis at home, this includes nursing services only and not care provided by carers or family members. This may be 16 hours, 24 hours or 32 hours.

Hours of day nursing care provided on a monthly basis at home, this includes nursing services only and not care provided by carers or family members. This may be 30 hours, 50 hours or 80 hours.

Hours of Physiotherapy provided on a monthly basis, this might be 1 hour, 2 hours or 3 hours.

Hours of Speech & Language therapy provided on a monthly basis, this might be 1 hour, 2 hours or 3 hours.

Days of respite out of home, provided on a monthly basis, this might be 1 day, 2 days or 3 days spent in a respite facility outside the home environment.

***Cash maintenance payment**, provided on a monthly basis, to go towards household expenses, this might be €50, €100 or €150. Please note this is not currently available and has been added for research purposes only

The services offered are COMBINED MONTHLY packages of care, provided by a mix of organisations including the JJE, HSE & other voluntary care providers.

Section 2:

This section asks you for some further information specific to you, such as your child's diagnosis, family size, additional financial expenses & other services.

SECTION 1

The 12 choice sets are presented below. Please select which monthly care package (A or B) you prefer most by putting a tick in the appropriate box below. Thank you.

Choice set 1	Monthly care package A	Monthly care package B
Night nursing hours	32 hours	16 hours
Day nursing hours	30 hours	80 hours
Physiotherapy hours	2 hours	2 hours
Speech & Language hours	2 hours	2 hours
Respite out of home days	2 days	2 days
*Cash maintenance	€100	€100

Choice A or B? (Please tick one)

Choice set 2	Monthly care package A	Monthly care package B
Night nursing hours	16 hours	32 hours
Day nursing hours	80 hours	30 hours
Physiotherapy hours	2 hours	2 hours
Speech & Language hours	3 hours	1 hour
Respite out of home	1 day	3 days
*Cash maintenance	€50	€150

Choice A or B? (Please tick one)

Choice set 3	Monthly care package A	Monthly care package B
Night nursing hours	24 hours	24 hours
Day nursing hours	80 hours	30 hours
Physiotherapy hours	1 hour	3 hours
Speech & Language hours	2 hours	2 hours
Respite out of home	2 days	2 days
*Cash maintenance	€150	€50

Choice A or B? (Please tick one)

Choice set 4	Monthly care package A	Monthly care package B
Night nursing hours	32 hours	16 hours
Day nursing hours	55 hours	55 hours
Physiotherapy hours	3 hours	1 hour
Speech & Language hours	3 hours	1 hour
Respite out of home	1 day	3 days
*Cash maintenance	€100	€100

Choice A or B? (Please tick one)

Choice set 5	Monthly care package A	Monthly care package B
Night nursing hours	16 hours	32 hours
Day nursing hours	30 hours	80 hours
Physiotherapy hours	3 hours	1 hour
Speech & Language hours	1 hour	3 hours
Respite out of home	1 day	3 days
*Cash maintenance	€150	€50

Choice A or B? (Please tick one)

Choice set 6	Monthly care package A	Monthly care package B
Night nursing hours	24 hours	24 hours
Day nursing hours	55 hours	55 hours
Physiotherapy hours	3 hours	1 hour
Speech & Language hours	1 hour	3 hours
Respite out of home	3 days	1 day
*Cash maintenance	€150	€50

Choice A or B? (Please tick one)

Choice set 7	Monthly care package A	Monthly care package B
Night nursing hours	16 hours	32 hours
Day nursing hours	30 hours	80 hours
Physiotherapy hours	1 hour	3 hours
Speech & Language hours	3 hours	1 hour
Respite out of home	3 days	1 day
*Cash maintenance	€150	€50

Choice A or B? (Please tick one)

Choice set 8	Monthly care package A	Monthly care package B
Night nursing hours	32 hours	16 hours
Day nursing hours	55 hours	55 hours
Physiotherapy hours	1 hour	3 hours
Speech & Language hours	2 hours	2 hours
Respite out of home	1 day	3 days
*Cash maintenance	€100	€100

Choice A or B? (Please tick one)

Choice set 9	Monthly care package A	Monthly care package B
Night nursing hours	24 hours	24 hours
Day nursing hours	80 hours	30 hours
Physiotherapy hours	3 hours	1 hour
Speech & Language hours	3 hours	1 hour
Respite out of home	3 days	1 day
*Cash maintenance	€50	€150

Choice A or B? (Please tick one)

Choice set 10	Monthly care package A	Monthly care package B
Night nursing hours	16 hours	32 hours
Day nursing hours	80 hours	30 hours
Physiotherapy hours	1 hour	3 hour
Speech & Language hours	1 hour	3 hours
Respite out of home	2 days	2 days
*Cash maintenance	€50	€150

Choice A or B? (Please tick one)

Choice set 11	Monthly care package A	Monthly care package B
Night nursing hours	32 hours	16 hours
Day nursing hours	55 hours	55 hours
Physiotherapy hours	2 hours	2 hours
Speech & Language hours	1 hour	3 hours
Respite out of home	2 days	2 days
*Cash maintenance	€50	€150

Choice A or B? (Please tick one)

Choice set 12	Monthly care package A	Monthly care package B
Night nursing hours	24 hours	24 hours
Day nursing hours	30 hours	80 hours
Physiotherapy hours	2 hours	2 hours
Speech & Language hours	2 hours	2 hours
Respite out of home	3 days	1 day
*Cash maintenance	€100	€100

Choice A or B? (Please tick one)

I have filled in all of the 12 choice sets

Are there any services that you particularly value that are not mentioned in the questionnaire above? Please provide some comments.

Now that you ticked a choice in each of the 12 scenarios, we are now interested in the actual amount of services you receive on a monthly basis and the amount you would like to receive. Please add any additional services that are not listed below:

	Current amount you receive	Amount you would like to receive
Night nursing hours		
Day nursing hours		
Physiotherapy hours		
Speech & Language hours		
Respite out of home		

How difficult did you find questions in Section 1: (Please tick box below)

- Not at all difficult
- Only moderately difficult
- Very difficult
- Extremely difficult

SECTION 2
FAMILY SITUATION

What is your child's diagnosis, if known?

What is your child's date of birth?

How many children do you have in total?

Do you have more than one child with a disability? If yes, what is your child's diagnosis?

What is your current marital status?

- Married
- Living with partner
- Single
- Separated
- Divorced
- Widowed

We are interested in studying the difference between the location parents live in and the variation in services they receive. Please provide your living location below.

Town :

County:

Who is the person providing most of the daily care for your child?

Did you or your partner / spouse have to give up work to care for your child? If so, can you please give an approximate loss of salary as a result?

What are you and your partner/spouse's occupations?

How long is your child receiving services at home?

Years:

Months:

**What is your Jack & Jill monthly budget allocation? What do you use this budget to pay for?
Please tick the box below.**

Nursing care:

Carer:

Family member (acting as carer):

ECONOMIC SITUATION

Are you in receipt of a carer's allowance?

Yes

No

Are you in receipt of a domiciliary care allowance?

Are you in receipt of carers benefit?

Do you have a medical card? If not, how long are you waiting for one?

We understand caring for a child with complex care needs has many additional financial costs. We have listed some examples below. Please fill in approximate costs and add other examples related to your situation. Please also include non-reimbursed expenses (yet to be paid):

Service type:	Cost:
Private Physiotherapy / other services	
Equipment for your child	
Travel costs (fuel + accommodation + babysitting costs)	
Electricity bills / oil bill / heating bill	
Phone bill	
Medication costs	
Adaptations to home / car	
Non reimbursed expenses (yet to be paid):	
Other:	

We would be most grateful if you could provide an estimate of your weekly household income from all sources (after tax and including your partner / spouse). PLEASE EXCLUDE ALLOWANCES (e.g., carer's allowance). We would like to remind you that the information you provide will be treated in the complete confidence. Please tick the appropriate box below.

- Less than €50.00 per week
- €50.00 – €100.00 per week
- €101.00 – €250.00 per week
- €251.00 – €500.00 per week
- €501.00 – €750.00 per week
- €751.00 – €1,000.00 per week
- €1,001 – €1,500 per week
- €1,501 and greater

OTHER SERVICES

Where do you get information about services for your child?

- JJF Liaison nurse
- Facebook
- Word of mouth
- Public Health Nurse

Other:

On average how many health care professionals visited your home in June 2013? Please fill out the box below and add any other additional health care professionals not listed.

Health care worker	Monthly visits e.g. 1 – 5 visits per month
Public health nurse	<hr/>
Early intervention nurse	<hr/>
Disability liaison nurse	<hr/>
Outreach nurse	<hr/>
Visiting teacher	<hr/>
GP	<hr/>
Liaison Nurse	<hr/>
Physiotherapist	<hr/>
Occupational Therapist	<hr/>
	<hr/>
	<hr/>

Many parents expressed an interest in psychological support services i.e., counselling. Would you and your partner avail of this service if it was offered to you? (Tick appropriate boxes)

- Yes, I would
- No, I am not interested
- My partner would
- My partner would not

Do you currently use a counselling service?

If yes, how much does this cost?

If no, why do you not use this service?

Do you think this service should be offered as part of a care package?

If you had to pay for a counselling service, how much would you be willing to pay for this service?

If offered, would you use 2 hours of your Jack & Jill monthly allowance to pay for a counselling service for you or your partner?

Finally, please provide any feedback or comments that you feel may be relevant to this study below:

WE VERY MUCH APPRECIATE THE TIME AND EFFORT YOU HAVE PUT INTO ANSWERING THESE QUESTIONS AND WE ASSURE YOU THAT ALL YOUR ANSWERS WILL REMAIN CONFIDENTIAL.

**PLEASE RETURN YOUR COMPLETED QUESTIONNAIRE
TO US IN THE PRE-PAID ENVELOPE PROVIDED by
FRIDAY 12th of JULY 2013. THANK YOU.**

Aoife McNamara & Professor Charles Normand
Department of Health Policy & Management
Trinity College Dublin



TRINITY COLLEGE DUBLIN
COLÁISTE NA TRÍONÓIDE, BAILE ÁTHA CLIATH

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OF DUBLIN



Appendix 14 - DCE Checklist

Checklist for assessing quality of DCE (Lancsar & Louviere 2008)

Checklist	This study
1. Conceptualising the Choice Process	<ul style="list-style-type: none"> • Choice tasks used • Binary (Package A or B) • Forced choice – no opt out, not realistic. • Generic (unlabeled)
2. Attribute Selection	<ul style="list-style-type: none"> • Meeting (1) with 11 nurses from voluntary organisation. • 24 Semi-structured interviews were used to inform attributes. • Meeting (2) with 11 nurses to confirm interview findings. • Likert scale conducted with (N=5) families to refine list of services. • Cash payment (continuous variable) was included to estimate WTAC.
3. Level Selection	<ul style="list-style-type: none"> • Semi-structured interview data used. • Voluntary organisation data to inform nursing levels. • Three levels were assigned for each attribute yielding $3^6 = 729$. • Level ranges were realistic but broad to ensure trade-offs. • Levels were evenly spaced.
4. Experimental Design	<ul style="list-style-type: none"> • A combination of 729 was not feasible, so a fractional factorial (FF) design was used. • FF main effects design with 24 hypothetically designed packages. • Profiles generated and allocated using Ngene software.
5. Questionnaire Design	<ul style="list-style-type: none"> • Introductory letter, with appropriate level of background and contextual information, was provided. • Clear instructions were provided. • Words and colour were the medium used to communicate attribute/level information.

Checklist	This study
6. Piloting	<ul style="list-style-type: none"> • Pilot study conducted with: • N=4 Families • N=4 Researchers • N=1 Expert • N=2 Nurses
7. Population / Study Perspective	<ul style="list-style-type: none"> • Families caring for children with neurodevelopmental life-limiting conditions. • Health services research perspective
8. Sample and Sample Size	<ul style="list-style-type: none"> • Targeted relevant population • Sample size calculated using ORME's Margin of error using 95% CI.
9. Data Collection	<ul style="list-style-type: none"> • Postal questionnaires with stamped addressed envelopes
10. Coding of Data	<ul style="list-style-type: none"> • Dummy variable coding
11. Econometrics Analysis	<ul style="list-style-type: none"> • Logit model • Mixed logit model • Latent Class model • Interaction effects of income and location.
12. Validity	<ul style="list-style-type: none"> • No answers were deleted • External validity obtained through presenting the results to experts. • The signs on the estimated parameters were consistent with <i>a priori</i> expectations.
13. Interpretation	<ul style="list-style-type: none"> • Results were in line with prior expectations and confirmed by using a mixed methods study.
14. Welfare and Policy Analysis	<ul style="list-style-type: none"> • Marginal willingness to accept compensation (WTAC) used to estimate monetary valuations. • Simulations and predicted uptake. • Probabilities of selecting packages based on attributes.

Appendix 15 – Free Text Comments

Discussion	Theme	Comments
<p>The following three comments provide further evidence on the concerns parents have over services for child after four years of age. Parents plan months in advance to try ensure services but receive no response. The uncertainty over care in the future is a major source of anxiety for parents.</p>	<p>Future concerns</p>	<p><i>“The sense of uncertainty now that the child is over four years cannot be overstated. Losing the JJF service was a huge blow”</i></p> <p><i>“Recommendations from JJF were sent 9 months prior to JJF withdrawal. It simply sat on a manager’s desk. This is a huge stress for parents, which is not needed.”</i></p> <p><i>“As my son turns 4 in October, I am very anxious about the help/support the HSE will give us when we no longer have JJF. We need as much help/support as is available to help care for our son at home.”</i></p>
<p>The need for counselling is highlighted in the preceding comments provided where parents where they reflect on the need for counselling supports to be provided at the beginning. One of the parents explained that had they received counselling, it may have saved their marriage.</p>	<p>A need for counselling</p>	<p><i>“At the beginning, just a few weeks after having our child, looking back, we both desperately needed counselling.”</i></p> <p><i>“We as parents believe the first service that should be offered in a care package being provided for a child with complex needs, is a counselling service for parents. In order to be able to care and support your child as a parent must be able to cope and adjust to the emotions, shock and upset a diagnosis brings. The mental health of parents is vitally important when caring for a child with complex care needs. This benefits all children in the</i></p>

<p>Slow referral was again mentioned as a serious problem with this particular family waiting 2.5 years for services to be set up.</p>	<p>Slow referral</p>	<p><i>family.”</i></p> <p><i>“My child was so well looked after over the past few years that somewhere along the way his parents lost each other and are now separating. We were so caught up in looking after him ... we spent no time together as a couple ... just maybe if we had someone (professional counsellor) to call to us now and again to make us realise how important it was for us to discuss and get through such unexpected circumstances surrounding the birth of our child.”</i></p> <p><i>“We knew our child had CP from day 2, but we were only put into contact with the JJF when he was 2.5 yrs. It would have made things much easier at the beginning. I think families should be referred to JJF much earlier, when we did get the JJF help, it has such a positive impact on our family.”</i></p>
<p>Battling for services is and the length of time for service set up is ...</p>	<p>Battle with services</p>	<p><i>“I wish we had known about JJF services earlier when (child’s name) was born.”</i></p> <p><i>“The HSE needs to change, it’s taken me 18 months of pure fighting to get nursing care services for my children” (Has two children with complex needs).</i></p>
<p>Parents describe that being new to the situation,</p>	<p>Information needs</p>	<p><i>“The difference between JJF and the HSE is that there’s better</i></p>

the lack of one source of information is overwhelming. When parents get connected to the JJF, they Liaison nurses serve as a primary source of information for parents.

One family calls for more investment into early intervention and that being refused additional services as a result of their partner working was unfair. Financial hardship was evident here where they described being overdrawn at the end of the month.

Financial challenges

use of money. What I feel is hard as a parent new to this situation is there is no one who can provide you with the information you need."

"Without JJF, we would know very little about most of the services etc. that are out there and find that there is little or no communication between all the different services and departments. My daughter sees four different people from the HSE and no two have the same name and address for her."

"It will be good if the government could invest more in early intervention. It is very difficult to get services like physiotherapy and occupational therapy which we need a lot. Considering experiences from this family, it is unfair that we were refused all other support because my partner is working. I would have got more if I was alone ... e.g. medical card. For the past four months, my husband is €2,500 overdrawn at the end of the month."

"We were paying a child-minder to care for our son in our home while we both worked but we have found this too difficult to maintain due to the financial strain. It would have been great to receive some financial assistance to pay this person as it was for five days a week."

One parent describes needing more of occupational therapy and physiotherapy and that they are needed to improve their child's condition.

Service needs

"We could really do with more O.T and Physiotherapy, we get some 2/3 times a month with Enable Ireland. But our child really needs more, as it seems to take very long to move forward. We feel if he has more services, he would improve, develop and make progress more quickly."

One parent describes using the JJF service to catch up on sleep and how they would be spending more time in hospital were they not in receipt of the JJF service.

Respite needs

"A full night's sleep is hard to come by so every now and then we need to use our monthly allocation of nursing hours to catch up on sleep. If we did not have JJF service, I am sure that my son would be spending a lot more time in hospital, and I am just as certain that either myself or my husband would have been in hospital at some stage too."

Managing the care of a child at home as described by one parent, changes family life dramatically.

Transition from hospital to home and impact on family

"Hospitals spend so much money on saving lives of children with severe complications, parents are sent home with their child with no supports (psychological, nursing etc.) or help. Life at home with other children and a partner changes dramatically. Impact on life with a disabled child (physical and intellectual) is huge."

One parent describes hearing about cutbacks on social medical very stressful and how it can cause more uncertainty around the services their child receives.

Stress of caring and social media

“Surely (at any cost) society should also be prepared to support a life when they leave hospital care. It is very stressful to listen to media reports on cutbacks to medical cards and supports (domiciliary & educational). I hope this survey helps in highlighting quality / quantity of support for families of children with disabilities.”

One parent describes the geographical problems with accessing care as a result of living outside the Dublin area. The quality of care is important to this parent and they want to be able to trust that their child is receiving the best care possible.

Out of home respite care, quality and geographic location

“Would love out of home respite care but would have to be high quality of care like for example Suzanne House, in Tallaght. I have had a bad experience with another respite facility so while out of home respite would give me an opportunity to concentrate on my child’s siblings and relax and catch up on sleep or visit family and friends down the country. I would not accept respite without a very high standard of care. My child’s progress is very slow so monthly speech and language therapy and physio with home programs or attending group physio sessions with home respite works really well. OT service except for equipment is a huge void in care. There are no end of geographical disadvantages because we live outside the Dublin area.”

<p>One parent describes the role of the JJF as fundamental to helping them cope with their child's illness.</p>	<p>Support framework</p>	<p><i>"Although we no longer use the JJF service, it was invaluable to us in 2011 and JJF got me back on my feet and helped me cope with the issues. It has helped us as a family to accept we needed help and to set up the framework for actually helping our child. She is now benefitting from this intervention. Thankfully we were able to stop using the funding but without it initially we would not have survived."</i></p>
<p>A parent talks of the long waiting times they experience in the provision of equipment and the fact that some of it is not funded.</p>	<p>Equipment</p>	<p><i>"As regards to the costs, these are paid by the health service, but some equipment takes at least half a year to get. Some equipment I need for son right away. Some equipment not funded. I am not able to get the finance myself. For example bathing aid, stair lift".</i></p>
<p>Trust was an important element for this parent, they suggest a more regulated system where health care staff sign in and out so there is a more transparent system in place.</p>	<p>Trust</p>	<p><i>"It is very easy for a cash payment to pay for a carer/friend/family member. It's based on trust, another method of delivery of payment should be brought in, to ensure that the correct service is received by the child. Maybe the carer should sign in and out per day/hours of care given. Sign in sheets posted to JJF."</i></p>
<p>The Liaison nurse is a highly valued service by parents. The Liaison nurse provides a broad extensive role in the supporting families and this is very much valued by this parent.</p>	<p>The essential role of the liaison nurse</p>	<p><i>"JJF have saved my family. From the minute I met the Liaison nurse, she was so positive. She completely believed in me. She helped me fight for my son's homecare package. We had been in Temple Street for 6 months with no end in sight. My Liaison nurse also arranged affordable counselling for me. She sorted entitlements when I wasn't capable of anything. Life is so much</i></p>

The intensive nature of caring for children with LLC's is described by one parent as relentless and demanding. They describe the lack of suitable services available to provide and the lack of practical information available.

Intensive nature of caring and lack of services available for children with complex care need

better now. I am very grateful for the support i receive. I believe this whole experience I receive, It has made me a better person.”

“Our child does not have huge medical needs at present. However, when he was first diagnosed he cried constantly, would only sleep for 20 mins at any one time and would take 1.5 hours to feed every three hours for the first year of his life. Our life was shattered and the state does not provide any minding facilities for children with complex needs. The private crèche wouldn't take him and none of the health care workers were giving us any practical information on how we might be able to find somebody to care for our son during the day to allow us some normal life.”

Appendix 16 - Oral Presentations and Posters

Year	Title	Conference / Venue
2012*Oral Presentation	<i>"A comparison of costs and outcomes of alternative models of care for children with complex disabilities in Ireland"</i>	Conference (AIRC): Optimising Health in the 21st Century, Trinity College Dublin; 7th - 8th November 2012
2013*Oral Presentation	<i>"Comparing the costs of providing care to children with life-limiting disabilities in Ireland"</i>	32 nd Annual International Nursing and Midwifery Research & Education Conference "Nursing in times of Austerity" hosted by the Royal College of Surgeons (RCSI) Dublin (2013)
2013*Poster	<i>"Eliciting parents' preferences of care services for children with complex care needs – Pilot results from a discrete choice experiment"</i>	Child Health Research - The key to a healthier European Society: (2013) Dublin City University
2013*Invited Seminar	<i>"Using discrete choice experiments in health services research"</i>	CHEPA – Centre for Health Economics & Policy Analysis, McMaster University, Ontario, Canada (Dec 2013)

2013*Oral Presentation Department seminar	- <i>“Interviewing families caring for children with life-limiting disabilities – study design & preliminary findings”</i>	Department of Health Policy & Management, School of Medicine, Trinity College Dublin. (May 2013)
2014*Invited Talk	<i>“Preferences of community care services for children with life-limiting disabilities - A novel way to elicit preferences”</i>	CAN CHILD: Centre for childhood disability research, McMaster University, Ontario (Feb 2014)
2014*Oral Presentation	<i>“Preferences of care services for children with life-limiting disabilities in Ireland – A discrete choice experiment”</i>	iHEA and ECHE Joint Congress: Health Economics in the Age of Longevity hosted by the Department of Health Policy & Management (2014) Trinity College Dublin
2014*Oral Presentation	<i>“Preferences of community care services for children with life-limiting Neurological Disabilities in Ireland - A discrete choice experiment”</i>	20 th International Congress on Palliative Care: Special Seminar on Pediatric Palliative Care, Montreal Canada. Sept 9 th -12 th (2014)

Extracurricular courses completed during PhD

- A short intense evening course in Basic Applied Statistics using R statistical software (TCD, May - June 2010) Dr. Allen Kelly)
- Topics in Microeconometrics (NUIG, July 2011, Professor William Greene)
- Applied Workshop: Using Discrete Choice Experiments in Health Economics: Theoretical and Practical Issues (Banff, Alberta, Canada. April 2012, Prof.Mandy Ryan)
- Introduction to Grounded Theory (DCU, June 2012, Prof Michael K.Thomas)
- Discrete Choice Modeling (NUIG, July 4-6th 2012, Prof.William Greene)
- Statistical Modelling in Epidemiology (TCD, Dec 2012, George Savva - TILDA)
- The Science of Behaviour and Mind - 18 week Psychology course (2013) (TCD)

HRB / OTHER Workshop Attendance:

- Effective Presentations for Postgraduate Research Students (2011) (TCD)
- The Publication Process (2011) (RCSI)
- Turbo Charge your Writing (2012) (UCC)
- Writing/Submitting your PhD & Preparing for your Viva (2012) (RCSI)
- Developing Critical Arguments for PhD Students (2012) (TCD)
- Systematic Literature Reviews (2012) (TCD)
- Completing Ethics Applications (2012) (RCSI)
- Research into Policy workshop (2012) (RCSI)
- Graduate Information and Literacy Skills Module (2013) (TCD)
- NVIVO workshops X 2 (2012 & 2013) (TCD & RCSI)
- Interpretative Phenomenological Analysis (2013) (TCD)

Research Travel Bursary Award

All Ireland Institute of Hospice & Palliative Care (AIHPC). International Research Summer Academy. Research Travel Bursary Award: April 2013

Attended summer academy at Lancaster University for weeklong course to develop social research methods relevant to Palliative and End of life Care.

Research Publications

1. McNamara-Howard A, Normand C, P. Preferences of community care services for children with life-limiting Neurological Disabilities in Ireland - A discrete choice experiment [Abstract]. In: Journal of Palliative Care - 20th International Congress on Palliative Care. Montreal Canada. (2014) p.199 Abstract: AO1-B.
 2. Reville, P., Ryan, P., McNamara, A., & Normand, C. (2013). A cost and outcomes analysis of alternative models of care for young children with severe disabilities in Ireland. ALTER-European Journal of Disability Research/Revue Européenne de Recherche sur le Handicap, 7(4), 260-274.
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Other:

3. McNamara, A., Normand, C., & Whelan, B. (2013) Patterns and determinants of health care utilization in Ireland. Report available on www.tilda.ie
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