‘It’s constantly having to push and fight, you feel physically and mentally drained’: A mixed methods exploration of the health and caregiving experiences of mothers and fathers of children with a life- limiting condition

Victoria Fisher

PhD

University of York

Health Sciences

March 2024

Abstract

Background: There are a growing number of parent caregivers providing extensive care for their child at home. We know little about how parents feel their own health should be supported. Fathers are underrepresented in the caregiving literature, meaning that we know very little about their experiences, health, and support needs and how their experiences compare to those of mothers.

Methods: A systematic review was conducted to identify and synthesise current evidence related to the experiences of fathers of children with a life-limiting condition. A qualitatively driven mixed methods study focused on fathers was then undertaken alongside qualitative interviews with mothers. Fathers completed an online survey comprised of demographics and self-report measures related to their health, sleep, and caregiving appraisals. They also took part in an optional semi-structured qualitative interview, to explore these issues in more depth. Basic descriptive statistics and reflexive thematic analysis were used to analyse the quantitative and qualitative data respectively. Semi-structured, in-depth interviews were conducted with mothers. Data was analysed using reflexive thematic analysis and used as a point of comparison for the fathers' data.

Findings: Thirty-two fathers took part in the survey, and twelve in interview. Fathers reported high levels of carer strain and distress, alongside high levels of family wellbeing and positive appraisals of caregiving. Compared with population norms, their sleep disturbance scores were high. Reflexive thematic analysis resulted in three themes: *'precarity in fathers' everyday lives', ' feeling understood in the context of health and wellbeing'*, and *'trajectory of child's illness; the importance of temporality'.* Thirty mothers took part in an interview. Mothers felt unable to prioritise their own needs, relative to those of their child and worried about who would look after their child if they did become unwell. They described stress as a result of battles with services rather than as a result of caregiving.

Conclusions: Fathers' extensive and overwhelming daily routines are inflexible and unstable, leading to cumulative precarities and a sense of role overload. Current provision is unable to address the unique and fluctuating support needs of mothers and fathers, which are inextricably linked to those of their child, and need to be understood in the context of parenting **and**

caregiving. A process capable of identifying and addressing parents' support needs in practice needs to be established.

Table of Contents

List of tables

List of figures

Acknowledgements

There are many people I would like to thank for their contributions to this thesis. First, I would like to thank the mothers and fathers who took part in this research. I am immensely grateful for the time they took to share their experiences with me so openly. I would also like to thank the members of the Martin House Research Centre advisory panel for their input to this thesis and for their support and guidance over the course of my PhD.

I thank my supervisors Professor Lorna Fraser and Professor Karl Atkin, for their patience, support, and encouragement, and for helping to build my knowledge and confidence as a researcher. I also thank Dr Jo Taylor for supervising me during the first year of my PhD, for continuing to take an interest in my research, and for offering words of support and encouragement when needed. I thank my Thesis Advisory Panel members, Dr Sarah Blower and Professor Fliss Murtagh, who have provided highly valued insight and support throughout my PhD. Thank you to Professor Kate Flemming who chaired my progression meeting. I would also like to thank all my colleagues and fellow PhD students within the Department of Health Sciences, for cultivating such a wonderful and inspiring research environment. Thank you to Dr Julia Hackett for supporting my study whilst I was on maternity leave.

Last but not least, I thank my friends and family. To James, my mum and dad, and Meg, for encouraging, supporting, and believing in me. To Theodore Atlas, thank you for bringing us so much joy, weekends will look a little different from now on!

Author's Declaration

I declare that this thesis is a presentation of original work and I am the sole author. This work has not previously been presented for an award at this, or any other, University. All sources are acknowledged as referenced.

Parts of this thesis have been disseminated in the following publications and presentations:

Publications

Fisher V, Fraser L, Taylor J. 2023. Experiences of fathers of children with a life-limiting condition: a systematic review and qualitative synthesis. *BMJ Supportive & Palliative Care*, 13, 15-26.

Fisher V, Atkin K, Fraser LK. 2022. The health of mothers of children with a life-limiting condition: A qualitative interview study. *Palliative Medicine*, 36(9), 1418-1425.

Fisher V, Atkin K, Ewing G, Grande G, Fraser LK. 2024. Assessing the suitability of the Carer Support Needs Assessment Tool (CSNAT-Paediatric) for use with parents of children with a life-limiting condition: A qualitative secondary analysis. *Palliative Medicine*, 38(1), 100-109.

Conference presentations

Health Sciences Research Showcase, University of York	Oral presentation: The health and caregiving experiences of fathers of children with a life-limiting condition: a mixed-methods study	2023
18th World Congress of the European Association for Palliative Care in Rotterdam	Poster presentation: The health of fathers of children with a life-limiting condition: a qualitative study	2023

Health Sciences Research Showcase, University of York	Poster Presentation: The health of parents of children with a life-limiting condition	2022
Martin House Research Centre Biannual Conference, University of York	Oral presentation: experiences of fathers of children with a life-limiting condition	2021
17th World Congress of the European Association for Palliative Care Online	Poster presentation: experiences of fathers of children with a life-limiting condition: systematic review and qualitative synthesis ***Prize: top 3 poster abstracts in category***	2021

1. Introduction

This thesis reports a mixed-methods investigation into the health and experiences of mothers and fathers of children with a life-limiting condition. This chapter provides background to the thesis, including the definition and prevalence of life-limiting conditions in children, the impact on their parents, the inconsistent means of support available to these families, the dearth of evidence that exists in relation to fathers, and why this needs to be addressed.

1.1 Life-limiting conditions in children

Life-limiting condition is term often used to describe both life-threatening and life-limiting conditions; the former being conditions for which there is a no hope of a cure and from which a child will die prematurely, such as Duchenne Muscular Dystrophy, and the latter conditions for which curative treatment may be possible but can fail, such as cancer (Fraser et al., 2020b). Life-limiting and life-threatening conditions fall broadly into one of four categories (i) conditions for which treatment is feasible but can fail; (ii) conditions for which a premature death is inevitable; (iii) a progressive condition for which there is no feasible cure; (iv) irreversible but non-progressive conditions for which the likelihood of premature death is increased (TfSL, 2018). Hain et al (2013) developed a directory of 376 life- limiting/threatening conditions, demonstrating diversity in both diagnosis and trajectory. This adds a certain complexity in trying to understand how this population might be supported, though there is overlap in the way in which these 376 conditions present and therefore in the support needs of children and families. For the purposes of this thesis, 'life-limiting condition' will be used to encompass both life-limiting and life-threatening conditions.

Category 1

Life threatening conditions for which curative treatment may be feasible but can fail

Includes conditions such as cancer and irreversible organ failure.

Category 2

Conditions where premature death in inevitable.

Treatment is aimed at prolonging life and includes conditions such as Duchenne muscular dystrophy.

Category 3

Progressive conditions without curative treatment options.

Treatment may extend over many years and includes conditions such as batten disease and mucopolysaccharidoses.

Category 4

Irreversible but non-progressive conditions causing severe disability, leading to susceptibility to health.

Complex needs mean that children are at high risk of life-threatening events or health complications. Includes cerebral palsy.

Figure 1; categories of life-limiting categories as defined by Together for Short Lives (TfSL, 2018).

Although each individual life-limiting condition is rare, latest figures for life-limiting conditions in children show a prevalence of 66.4 per 10,000 in England in 2017/18, which is estimated to increase to between 67.0 and 84.22 per 10,000 by 2030 (Fraser et al., 2020b). Improved medical treatments and survival mean that these children may live with their condition for many years, often into adulthood (Jarvis et al., 2021). Many of these children have extensive healthcare needs such as mobility and ventilation support, gastrostomy feeding, and multiple medications either for the direct treatment of their condition or for symptom management (Fraser et al., 2020b). The unpredictable and diverse range of symptoms experienced by these children include pain, seizures, muscle spasms, communication issues and breathing problems, resulting in the need for high levels of intervention and frequent medical appointments (Jaaniste et al., 2021).

Data tells us that the prevalence of life-limiting conditions is greater in boys than girls (72.5 per 10,000 vs girls 60.0 per 10,000), is highest among children under 1 years old (226.5 per 10,000), and in those of Pakistani origin (103.9 per 10,000). It is also higher in children living in areas of highest deprivation (Fraser et al., 2020a). The most common diagnoses are congenital abnormalities i.e. structural or functional abnormalities present at birth. Although this accounts for the high prevalence of children under 1 with a life-limiting condition, it is important to reiterate that many children live beyond this, often into adulthood.

1.2 Care for children with a life-limiting condition

Children with a life-limiting condition will encounter a range of services over the course of their lives, which will change in accordance with fluctuating needs related to factors like age and stage of illness. Paediatric palliative care is an important component in the care of children with a life-limiting condition (Taylor et al., 2020) and is defined by the World Health Organisation (WHO) as "'the active total care of the child's body, mind and spirit . . . [that] begins when illness is diagnosed, and continues regardless of whether or not a child receives treatment directed at the disease" (WHO, 2011). This distinguishes paediatric palliative care from that of adults; paediatric palliative care is an approach to care, rather than an end-of-life phase of treatment (Mitchell et al., 2021).

There are various forms of palliative care services available to children, from those provided in the community, to specialist palliative care which is most often delivered in children's hospices or acute settings (Mitchell et al., 2021). In the UK, there are 54 children's hospice organisations that provide support to children and their families, either in a hospice or at home. Such services have the potential to take substantial pressure away from hospitals, reducing intensive care admissions, improving the quality of life for children and their families (Mitchell et al., 2019a) as well as meeting their preferences for place of death (Gibson-Smith et al., 2021). However, geographical differences mean that there is significant inequity in availability and access to support for individual children (Mitchell et al., 2017). Further issues such as delays in referrals mean that even children with the most complex needs do not have access to specialist services (Mitchell et al., 2019a, Mitchell et al., 2021). NHS England have previously announced grant increases for children's hospices in 2022/23 and again in 2023/24 but it is not yet clear whether such increases will be extended beyond this, meaning that hospices will have to make difficult decisions surrounding cuts to the care that they offer including respite, end of life care and symptom management support (TfSL, 2023).

Aside from specialist services, strains on primary and secondary care, including general practitioners (GPs) and community nursing teams, mean that families may be also missing out on vital support at home (Mitchell et al., 2019a). Whilst there is a growing body of evidence surrounding the experiences and needs of families, as well as the benefits of paediatric palliative care, the implications for policy have been limited, particularly in terms of how services are able to meet growing demands of a heterogenous population.

1.3 Impact of caregiving on parents

The parents of children with life-limiting conditions commonly provide all their child's care at home which, as mentioned, can be extensive, can come with varying levels of support, and can extend over many years (Mitchell et al., 2019a). Some parents may also have to manage their child's symptoms alongside the uncertainty of the lack of a formal diagnosis and undefined prognosis for their child which is so often the case for very rare conditions. Being able to meet the medical needs of their child can be daunting for parents who must learn to manage new equipment, administer medicines, and closely monitor their child's condition and symptoms (Verberne et al., 2017). In addition to meeting these medical needs, parents manage extensive appointment schedules, advocate for their child in education and healthcare settings and carry out extensive research surrounding their child's condition and treatment to improve knowledge and inform their decision making. This occurs alongside more general parenting responsibilities, and meeting the needs of other family members (Koch et al., 2021).

Research tells us that providing this level of care, alongside the knowledge that their child will die, can have a deep impact on parents' emotional, psychological, and physical wellbeing (Pinquart, 2018, Koch and Jones, 2018). Whilst individual responses vary, many parents will be left susceptible to mental and physical health difficulties. Studies demonstrate higher levels of stress, anxiety, depression, and stress disorders in these parents than in the general population (Collins, 2020, van Warmerdam et al., 2019, Pinquart, 2018). Parents have described a sense of exclusion from the 'outside world', partly pertained to a lack of time and energy to engage with anything other than caregiving which is an all-encompassing task, physically and mentally dictating their schedules (Postavaru, 2018). Loneliness and isolation have been well cited in research studies spanning the last 30 years (Oakley et al., 2021). As well as the loss of social connections, and the opportunity to maintain relationships, some parents can also be excluded from the workforce, and experience a decline in income leaving them with financial problems (Pelentsov et al., 2015). Families may also experience financial problems related to extra costs associated with equipment for their child (Mitterer et al., 2021). Low-income families are particularly vulnerable to the effects of reduced income and costs of caring (Avery et al., 2024).

Complex and often conflicting emotional states can exist simultaneously for these parents. Parents have described experiences of grief, sadness, fear, anger and guilt, alongside a real sense of hope and determination in managing their child's condition (Postavaru, 2018, Gill et al., 2020), personal growth, empowerment, a new found spirituality, and developing new and important relationships with other parents through shared understanding (Gérain and Zech, 2019). It is evident that parents need to be appropriately supported to provide care for their child, particularly when we consider the extent of care that many of these families provide for extended periods of time. However, understanding what 'appropriately' means in this context is not straightforward and there is no standard means of support available to parents. Reviews have identified parents most frequently cited needs spanning informational, physical, psychological, emotional, social, and practical domains (Pelentsov et al., 2015, Gill et al., 2020) but as mentioned, there still remains a significant gap between research and meaningful changes to practice. We do know that optimal support needs to be able to address the needs of individuals within the heterogeneity of the population, but understanding what this support might look like is made difficult as samples in research studies continue to be relatively homogenous. Furthermore, studies have rarely addressed parents' own health needs directly. As referenced above, there have been several epidemiological studies surrounding mental health outcomes in parents, and particularly mothers, but such have not been explored qualitatively nor have parents been asked how they want their health needs to be addressed.

Another key limitation of the existing body of research is the lack of inclusion of the perspectives of fathers. Mothers make up the majority of studies focused upon parental experience and so we know little about the broader experiences of fathers as well as how they want to be supported. There is a need to understand whether this differs from the support needs of mothers, and how such can be incorporated into the delivery of care and support.

1.4 The caregiving father

Historically, the centrality of the mother-infant attachment has seen fathers and other family members on the periphery of caregiving (Lamb, 1975). Fathers have, cross culturally, been seen as the 'breadwinners', responsible for taking care of their families financially (Novianti et al., 2023). Contemporary family dynamics represent a shift in this role dichotomy, with fathers increasingly expected to be involved in their child's care and development (Diniz et al., 2021). However, these changes exist alongside enduring social attitudes that are reminiscent of those traditional perceptions of parenting roles, creating barriers to some fathers being able to truly achieve greater involvement with their children (Petts, 2022). Uptake of extended paternity leave, or shared parental leave, is rather small (Kaufman, 2018), and the gender pay gap remains a central issue in families being able to achieve financial stability if mothers want to return to work (Newton et al., 2018). This is clearly a broad and complex issue, requiring more attention than the scope of this thesis can allow. However, societal expectations of parenting roles have specific implications for families of children with complex needs, both in research and in practice. One parent will *usually* take the bulk of the caregiving, whilst the other goes to work, but the way in which research has approached this topic means that the caregiving contributions of the employed parent, usually fathers, can be undermined (Tan et al., 2019).

1.5 Paternal involvement in research

There is a distinct shortage of information surrounding the experiences and support needs of fathers of children with a life-limiting condition. Studies reporting the experiences of parents of children with a life-limiting condition often include very small sub-samples of fathers meaning that findings and subsequent recommendations for policy or practice, including the aforementioned studies, are based upon the view of mothers (Nicholas et al., 2020). This underlying bias in individual studies is especially noticeable in reviews of parental experiences, which often include numerous mother-only studies. For example, a meta-ethnography of the experiences of parents of children with a life-limiting condition (Postavaru, 2018) included 17 studies; 6 of which focused solely on the experiences of mothers, 10 on those of both mothers and fathers and just 1 exclusively on the experiences of fathers. Furthermore, as recognised by the author, the mixed studies were heavily biased towards mothers (Nicholas et al., 2020). In the context of the experiences and needs of caregivers, this imbalance would once have been expected and accepted alongside the traditional dichotomy of parental roles, as mentioned above. However, evolving family dynamics, and contemporary understandings of the impact of childhood illness, call for studies that seek to address this imbalance to make sure that all family members are supported (Aoun et al., 2022).

When a child is diagnosed with a life-limiting condition, parents are expected to provide very extensive care for their child, often requiring swift adaptation to new roles and responsibilities (Koch et al., 2021). The demands of caregiving can be vast, meaning that parents may need to alter their perspectives and expectations of family life (Oakley et al., 2021). As mentioned, current ideas surrounding how best to support parents, rests mainly upon evidence from mothers, but if we are to truly embrace the family-centred nature of the palliative care model, we must look beyond this. The focus on mothers not only fails to capture important paternal views in the development of supportive interventions but overlooks the impact of caregiving on parental and familial relationships and vice versa. Understanding why some parents seem to cope relatively well with caregiving, whilst other may struggle, rests upon the consideration of factors at individual, familial, and societal levels (Raina et al., 2004). Including the perspectives of fathers in this discourse begins to address this, allowing us to form a more holistic view of the experiences and support needs of families, which in turn supports the care of the child.

Addressing the challenges of recruiting fathers to research has been the focus of many studies, both in this population and in parenting research more broadly (Nicholas et al., 2020, Yaremych and Persky, 2023). Despite some reorganisation of parental roles, mothers are still more likely to be labelled as their child’s primary caregiver, which has ongoing implications in research. Gender-based assumptions surrounding fathers’ willingness to share their experiences, mothers being more available to take part in research, and mothers knowing more about their child’s condition are heavily cited in relation to fatherhood research barriers (Nicholas et al., 2020). So, even when studies seek the views of mothers and fathers, mothers are inherently more likely to be approached by those recruiting, reinforcing gender-based tensions in research and in turn upholding the biases within policy and practice. This is further highlighted in studies that demonstrate that low participation is rarely due to lack of interest as opposed to fathers just not being asked to participate (Davison et al., 2017). Furthermore, some fathers **will** be their child's primary caregiver but have nonetheless been excluded from research.

Improving the inclusivity of research and paying more attention to the experiences of fathers specifically, is essential for understanding the needs of these fathers, whether these differ from those of mothers, how this contributes to our understanding of family support and in ensuring that mothers and fathers feel equipped to care for their child. Central to this thesis is the distinction between research that seeks to understand parental perspectives of their child's condition and care, and research that seeks to understand parents' *own* experiences and needs associated with having a child with a life-limiting condition. There is of course overlap here, though highlighting the importance of parental perspectives despite level of involvement in, or their understanding of, their child's condition and care, is key to a greater understanding of family support needs. Research that develops upon what we know about mothers *and* fathers is warranted. Below I detail the outline of this thesis demonstrating how I have addressed gaps in the literature in relation to both mothers and fathers.

1.6 Outline, structure, and methodological approach of this thesis

In this **introductory chapter**, I have defined life-limiting conditions and detailed the prevalence of these conditions in children. I have explored current evidence relating to the impact of caregiving on the parents of these children, highlighting the vast amounts of expertise and care that they contribute. However, studies have often overlooked fathers' experiences meaning that we know relatively little about their perspectives. They are also often absent from prevalence studies meaning that we also know little about health outcomes in these fathers. I have examined some of the reasons for this, and why it needs to be addressed, forming the overarching rationale for this thesis. Alongside the clear dearth of research surrounding fathers, I have also highlighted that although we know a little more about mothers' health outcomes, there has been little exploration from mothers' own perspectives about such outcomes and their preferences for support.

In **chapter 2**, I present the findings of my systematic review and qualitative synthesis exploring current evidence related to the experiences of fathers of children with a life-limiting condition, through which it will become clear that there are substantial gaps in the literature surrounding the experiences of fathers as caregivers and impacts on their own health. I include a discussion of the limitations of the included studies and go on to describe how these findings gave rise to chapters 4 and 5; a primary study focused on the health and caregiving experiences of fathers of children with a life-limiting condition.

At **chapter 3** I detail a primary qualitative study exploring the health of mothers of children with a life-limiting condition, including their interactions with services and what they do to look after their own health, which had so far not been explored in detail in the context of this population of parents. This chapter is the result of a broader field of work that I was involved in during my PhD for which I detail my role, study findings and their relevance to this thesis. Although initial plans did not account for the inclusion of the maternal health study in this thesis, through my involvement, it became clear that the study provided insight, and the opportunity for important comparisons with data I was collecting on fathers.

This chapter is presented prior to my methodological rationale for the primary study on fathers as: (a) data collection and analysis for the maternal health study occurred during a similar timeframe to me designing the fathers' study; (b) it contributed to the study design of the fathers' study; (c) it helped me to develop research skills that contributed to all aspects of the fathers' study. Key comparisons with the fathers' data will be presented in the discussion in **chapter 6**, through which I will develop conclusions surrounding implications for practice, and how the support needs of both mothers and fathers may be adequately addressed.

Chapter 4 sets out my primary research aims in relation to fathers:

- To describe the physical and mental health and well-being of fathers of children with a life-limiting condition, using self-report measures.
- To describe the caregiving experiences of fathers both quantitatively and qualitatively.
- To qualitatively explore fathers’ perceptions of their own health, well-being, and caregiving experiences.

The chapter is a detailed account of the mixed methods study design, my rationale for using a combination of quantitative (survey) and qualitative (interviews) data collection methods. I detail my methodological approach with emphasis on the need for an exploratory and flexible study design, capable of gathering rich insight into the experiences of fathers. I consider ethical issues and my own positionality/reflexivity.

Chapter 5 details the findings of the study; (1) the quantitative survey using basic descriptive statistics; (2) findings of the reflexive thematic analysis of the qualitative data, outlining three major analytical themes. The quantitative and qualitative components are initially presented separately, and then as a joint display, through which the quantitative data provides additional context to the results of the interviews. I also explore whether any of the quantitative findings can be explained, or elaborated on, through the interviews.

In **chapter 6** I bring together and discuss the results of each of these threads in relation to the wider context whilst reconsidering concepts introduced in chapter 1; societal perspectives of parenthood, and how we understand the roles of both mothers and fathers in the context of paediatric palliative care. I explore how these findings relate to policy, practice, and research. I describe the key strengths and limitations of my study, and this thesis, detailing some key lessons and reflections. I conclude with a summary of the original contributions that this thesis makes to current understanding in this area.

1.7 A note on patient and public involvement (PPI)

The studies detailed in this thesis benefited from the input of a family advisory board (FAB) (that had been established in 2018), made up of parents and other adult family members of children and young people with complex healthcare needs. The group provided input at each stage of my PhD; during the design and analysis stages of the systematic review, maternal health study, and primary father's study. I first met with the group in-person in October 2019, to introduce myself and the research that I would be conducting. I particularly sought some early input from the fathers in the group, who felt that a fathers only study was extremely important. Following this first meeting, the COVID-19 pandemic meant that subsequent meetings were held online. Although this created some challenges, the meetings were well attended, including by some new members. The number of attendees varied at each meeting but 5 fathers were part of the group and provided input across my PhD. Specific input for each thread of work is detailed in the relevant chapters. I met with the group several times in person/on zoom but had an ongoing dialogue with 2 of the fathers outside of the meetings who were particularly keen to provide input and support activities like piloting the survey and topic guide.

2. The experiences of fathers of children with a life-limiting condition: A systematic review and qualitative synthesis

As highlighted in chapter 1, what is known about the perspectives of fathers of children diagnosed with a life-limiting condition is relatively small compared to mothers. Reviews focused on parental experiences, as well as those looking more specifically at support needs, include very few fathers, meaning that recommendations for practice have generally been developed from the views and experiences of mothers (Nicholas et al., 2020). We know little about how fathers' experiences may differ, warranting a study able to explore their unique perspectives. To comprehensively assess the existing research of fathers' experiences, I carried out a systematic review and qualitative synthesis. My aim was to identify and synthesise qualitative findings related to the experiences of fathers of children diagnosed with a life-limiting condition. Initial scoping searches identified studies focused solely on the experiences of fathers, and a qualitative synthesis of these studies would help to gain a deeper understanding of issues that mattered to fathers, as well as gaps in the literature. The objectives of this study were to: (a) systematically review current literature to identify qualitative studies that look at the experiences and perspectives of fathers of children with a life-limiting condition; (b) synthesise the findings of these studies to gain a deeper understanding of fathers' experiences and perspectives.

This systematic review and qualitative synthesis has been published (see Appendix 1):

Fisher V, Fraser L, Taylor J. 2023. Experiences of fathers of children with a life-limiting condition: a systematic review and qualitative synthesis. *BMJ Supportive & Palliative Care*, 13, 15-26.

Existing reviews were either focused predominantly on children with cancer (Jones et al., 2010, Polita et al., 2018) or included studies in which mothers' perspectives were sought. A recent meta-ethnography explored the experiences of fathers providing care for their child diagnosed with a life-limiting condition (Postavaru et al., 2021) but again included mixed samples, some heavily biased towards mothers.

2.1 Methods

This review protocol was registered with PROSPERO CRD42020167076 on 16th March 2020 (https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=167076).

Qualitative evidence synthesis (QES), qualitative meta-synthesis or qualitative research synthesis are interchangeable terms used to describe a range of methodologies related to the systematic review of qualitative research (Flemming et al., 2019). These methods collate the findings of individual studies to enhance understanding of a particular phenomenon, improve generalisability and identify shared and contradictory perspectives that may be missed in standalone studies (Flemming et al., 2019). Noblit and Hare (1988) refer to synthesis as "making a whole into something more than the parts alone imply". In health research, this may be evidence pertaining to the acceptability, feasibility or cost-effectiveness of an intervention or to more general explorations of the experiences of, for example, those diagnosed with a particular disease (Lewin et al., 2015). This evidence is able to contribute to the implementation of interventions, policy formation, identifying and developing future research priorities and preventing the unnecessary duplication of primary research (Soilemezi and Linceviciute, 2018).

2.1.1 Choosing an approach

There are many qualitative evidence synthesis methodologies, each exhibiting key similarities and differences, both in their approach to synthesis and in their theoretical foundations (Barnett-Page and Thomas, 2009). The type of review question was a key consideration in choosing an approach to synthesis; consideration must be given to whether questions seek to address specific aims such as the effectiveness of an intervention, or to explore the general experiences and perspectives of individuals. This review sought to explore the latter for which little was known; the experiences of fathers of children with a life-limiting condition. Therefore, thematic synthesis, inductive in nature (Barnett-Page and Thomas, 2009), was deemed to be suitable to explore this topic area. The parameters of the question were then developed in line with the SPIDER framework (Cooke et al., 2012).

Table 1; SPIDER framework inclusion criteria

Concept	**Criteria**
Sample	Fathers, step- fathers, adoptive fathers, long- term foster fathers, bereaved fathers
Phenomenon of **I**nterest	Having a child of any age with a LLC
Design	Interviews, focus groups, ethnography etc
Evaluation	Experiences and perspectives of having a child with a life-limiting condition
Research type	Qualitative research

As evident from the parameters described in table 1, an approach capable of identifying commonalities and differences within broad experiences and across a number of study designs and methodologies (broadly ‘qualitative’) was required. Furthermore, it was important to consider that the aims of the individual studies would not necessarily align with the aim of the review i.e. some research questions would be more specific than broadly exploring fathers’ experiences. Therefore, an approach capable of dealing with heterogeneous data was required. These factors are strongly tied to epistemological considerations i.e. the capability of a method in dealing with studies from a range of settings and theoretical positions.

Barnett-Page & Thomas (2009) discuss each qualitative synthesis approach with respect to its position on the spectrum of ‘subjective idealism’ to ‘naïve realism’. The former assumes that reality exists as human construction, whereas the latter is characterised by the belief of an objective reality in which things are perceived as they are. This consideration is recognised as being important, though the extent to which review teams take epistemology into consideration does vary (Booth et al., 2018).

The vast array of methods available for synthesising research sit at various positions on this continuum. For example, meta-narrative synthesis, critical interpretive synthesis and meta-studies are heavily informed by ‘subjective idealism’, underpinned by their emphasis on interpretation and the premise that no single objective reality will be found (Barnett-Page & Thomas, 2009). Meta ethnography and grounded theory, although similarly reliant on interpretation, seem to be influenced by ‘objective idealism’, that is; “a world of collectively shared understandings” (Barnett-Page and Thomas, 2009). Emphasis is placed on identifying commonalities between studies, and contradictions explained with respect to contextual differences. Thematic synthesis and narrative synthesis go further in their assumptions that findings are able to represent a shared reality. Therefore, thematic synthesis enables the inclusion of studies that sit at a variety of epistemological positions, whereas methods such as grounded theory or meta-interpretation are more strongly underpinned by epistemological considerations (Booth, 2016).

As mentioned, thematic synthesis generally assumes that findings of individual studies are able to represent a shared reality. Whilst this was important for identifying commonalities amongst fathers’ experiences, there is some debate as to whether creating new interpretations of data can lead to the context of individual studies being lost and the nature of qualitative research undermined (Barnett-Page and Thomas, 2009). However, thematic synthesis is underpinned not only by its ability to produce new concepts and hypotheses that are explicitly linked to the findings of primary studies, but by the transparent nature of each step that the researcher undertakes in order to get there (Thomas and Harden, 2008).

Related to this is the number and type of studies that are included. Rich and thick studies, i.e. studies with great conceptual and contextual detail respectively, make interpretative methodologies such as meta-ethnography appropriate. However, if a large number of studies are included, these methods may not be able to provide meaningful results. Generally, descriptive methods such as meta-studies, can handle a greater number of studies and are more suited to contextually thin data (Booth et al., 2018). However, recommendations as to the number of studies that each approach should include are flexible and study dependent (Soilemezi and Linceviciute, 2018). Initial scoping searches provide an indication of the richness and thickness and should be considered in combination with studies’ heterogeneity

(Booth et al., 2018). Initial scoping searches for this study suggested that data would range from descriptive to interpretative and explanatory for which thematic synthesis was appropriate.

Timeframe, resource and expertise are three closely related considerations that also helped me to select the most appropriate method for synthesis. Booth et al. (2018) highlight that these practical considerations should not, in isolation, have ultimate bearing upon selection, but help to determine the feasibility of a preferred approach. They consider that studies for which a greater level of theoretical engagement is required, such as meta-ethnography, longer timescales and greater levels of expertise may be required. A benefit of thematic synthesis is that it is based upon established techniques of thematic analysis which are accessible to all qualitative researchers and flexible according to the needs of individual studies (Nowell et al., 2017).

Different approaches to synthesis enable the production of results suited to different audiences and purposes. For example, methods such as thematic synthesis and narrative synthesis can be highly relevant to policymakers, whereas the constructivist nature of methods such as Grounded Theory often functions at a more conceptual level (Booth et al., 2018). This consideration is not only defined by the intended reach of a review at an individual/group/organisation level e.g. policy makers, healthcare providers, patients, the public, but also in terms of geographical reach.

In summary, I selected thematic synthesis due to its accessibility and improved guidance, its ability to synthesise heterogeneous studies from a range of epistemological positions and its suitability for exploring under-researched areas. It shares characteristics with both meta-ethnography, in that overarching concepts are translated from one study to another and with Grounded Theory due to its inductive approach (Barnett-Page and Thomas, 2009). To ensure transparency of reporting for all stages of the review process, the review was reported in accordance with Enhancing Transparency in Reporting the synthesis of Qualitative research (ENTREQ) guidelines (Tong et al., 2012).

2.1.2 Inclusion criteria

The inclusion criteria and search strategy were developed in line with the SPIDER framework as described in table 1 (Cooke et al., 2012). Studies were included if:

- They used qualitative research methods to explore fathers', step-fathers', adoptive fathers' or long-term foster fathers' experiences of having a child with a life-limiting condition diagnosed in childhood. This included bereaved fathers.
- ≥60% of the child population were diagnosed with a life-limiting condition as defined by the diagnoses listed in search strategy. Mixed samples were to be expected, especially when we consider that many life-limiting/life-threatening conditions are extremely rare, making truly homogenous samples difficult to achieve. Deciding on a cut-off sample % for inclusion was challenging; generally, it is accepted that a common-sense approach be applied, balancing the risk of including ineligible sub-samples with the risk of losing valuable insights (McKenzie et al., 2019). However, others prefer to define an un-ambiguous cut-off for inclusion (O'Donnell et al., 2016), which was the approach taken here.
- They used a mixed-methods approach as long as the qualitative data were reported separately and could be clearly extracted. This is an approach used in other published qualitative syntheses (Flemming et al., 2020, Woodman et al., 2016).
- They were published in English. Divergent findings exist as to the risk of bias of excluding studies published in languages other than English (Neimann Rasmussen and Montgomery, 2018). This discussion becomes even more complex when we consider the importance of language and meaning in qualitative research and how translation, an interpretive process in itself, may affect the validity of findings (van Nes et al., 2010). Not only can meanings be lost in translation, but there may be words, phrases or metaphors that don't have a direct translation to English (Regmi et al., 2010). On the other hand, important contextual information may be lost by excluding studies published in languages other than English (Walpole, 2019). Consideration must also be given to the resource available i.e. funding for translation (Soilemezi and Linceviciute, 2018), as translation is a time consuming and resource intensive task, especially in the case of qualitative research where there may be vast amounts of data

and contextual information to translate (Regmi et al., 2010). It was for this reason that only studies published in English were included. However, studies that had been published in another language, and later translated and published in English were included.

- Studies were included regardless of geographical location. As with the exclusion of non-English studies, excluding studies based on country may have resulted in the loss of valuable insights. Different healthcare infrastructures can often make it difficult to compare results across countries, though thematic synthesis was able to take contextual factors into account.

2.1.3 Exclusion criteria

Studies were excluded if:

- They included the experiences of other participants i.e. mothers or professionals. This was a key decision made during protocol development. The growing interest in parental experiences has led to a proliferation of studies that explore the views of mothers and fathers as a homogeneous sample (Laws, 2018). These studies provide valuable insights, though it is often difficult to distinguish data pertaining solely to the views of fathers (Bally et al., 2018). Furthermore, PPI members suggested that true representation of the experiences of fathers should be sought in the absence of other experiences/voices. The family advisory board includes parents of children who have a life-limiting condition and meet once a month to discuss various projects.
- Studies in which qualitative data was analysed quantitatively were excluded. However, the methods by which qualitative data can be quantified exist on a spectrum, making inclusion decisions less straightforward. Qualitative studies may use simple quantitative approaches such as frequency counts as a means highlighting particular findings i.e. the most prevalent theme. Qualitative content analysis is an example of this in that “data are presented as words and themes” with “the possibility to add information by performing some quantification in which sub-categories and categories are counted” (Bengtsson, 2016). Some go slightly further, for example employing measures of association between themes and participant characteristics. Scoring systems have also been used to demonstrate the strength of participants’

experiences (Fakis et al., 2014). These approaches can be valuable (Mays et al., 2005) and can also be used in the integration of qualitative findings in mixed methods reviews (van Grootel et al., 2020). This spectrum of methodologies means that studies in which there was some numerical data were not immediately excluded. The exclusion criterion was applied to studies in which qualitative data had been analysed solely with quantitative methods and for which there were limited qualitative results to code. Studies in which qualitative data had initially been analysed qualitatively, with quantitative methods applied later, were included if the data pertaining to qualitative analysis was easily extracted.

2.1.4 Search strategy

The search strategy was developed using terms for life-limiting conditions (Fraser et al., 2015), father and children, and a qualitative filter was applied. The SPIDER tool (Cooke et al., 2012) was used to define terms for each concept and advice was sought from an information specialist during initial development.

Table 2; Search terms for each concept in SPIDER framework

Concept	**Criteria**	**Search terms**
Sample	Fathers	Father, Dad, Step-father, Adoptive father, Foster father, Foster-father
Phenomenon of **I**nterest	Having a child of any age with a LLC	Child, Infant, Baby, Toddler, Kids, Boy, Boyhood, Girl, Girlhood, Minor, Schoolchild, Adolescence, Juvenile, Teenager, Paediatric, Young people, Young adult (Taylor et al., 2020) Life-limiting illness, life-limiting condition, life-threatening

		illness, life-threatening condition, life-limiting disease, life-threatening disease Terms for specific diagnoses (Fraser et al., 2015)
Design	Interviews, focus groups, ethnography etc	Qualitative research, interview, semi-structured, personal narrative, narrative, audio recording, field notes, key informant, Focus groups, experiences, views, perspectives, beliefs, attitudes Phenomenology, hermeneutics, discourse analysis, grounded theory, thematic analysis, narrative analysis, ethnography, case study, themes, conversation analysis, descriptive study, exploratory study, inductive, content analysis (Coventry et al., 2016).
Evaluation	Experiences and perspectives of having a child with a LLC	
Research type	Qualitative research	

I identified studies through searches of electronic databases MEDLINE, CINAHL, EMBASE, PsycINFO and Social Science Citation Index in March 2020, using a combination of MeSH, keyword and free-text terms. These databases were chosen as they span nursing, medical, psychology and social science literature, making them appropriate for the research topic. No date limits were set. The strategy developed in MEDLINE, was adapted for each database

(Appendix 2). I supplemented electronic searches with citation searching, searching reference lists of included articles and a Google Scholar advanced search for grey literature.

I initially tested searches for sensitivity and specificity in MEDLINE using target papers. Three of the target papers were not appearing in the search results, and so the strategy was adjusted. Further and more exhaustive terms were added to the initially fairly broad qualitative filter until target papers appeared in the search results. This is in keeping with recommendations that relate to the inclusion of a combination of broad based, thesaurus and free text terms for maximum retrieval of relevant studies (Shaw et al., 2004). However, there is currently no consensus regarding whether or not strategies should encompass all three, or whether a broad- based strategy is just as effective (Flemming and Briggs, 2007).

The challenge of defining optimum and sufficiently extensive search strategies in qualitative syntheses have been acknowledged for several years (Barroso et al., 2003). Furthermore, what is considered as ‘sufficiently extensive’ can vary in the context of individual syntheses (Mackay, 2016). For example, some studies may require a comprehensive strategy, reflecting quantitative review practices, whereas some authors consider the appropriateness of studies to be of greater importance than comprehensiveness, and utilise methods capable of incorporating sampling, such as realist synthesis (Booth et al., 2013). Sensitivity and specificity of searches may also vary between databases. For example, an extensive qualitative filter may be deemed unnecessary in CINAHL, where qualitative indexing is relatively good, but other databases may perform less well (Booth, 2016). This inconsistent indexing across databases, as well as use of non-standardised terminology across qualitative methodologies and low quality or absence of abstracts contribute to the complexities of locating studies. Whilst methods must remain systematic and explicit, searching for qualitative research can be a non-linear, iterative process in which searches change as understanding evolves (Harris et al., 2018).

2.1.5 Screening and selection

I imported the results of my searches to Endnote and removed any duplicates. I then uploaded them to Covidence for screening. Title and abstract screening was undertaken by myself and another reviewer (AC). Discussions were held after each 1000 studies had been screened and any disagreements were resolved through discussion. We then reviewed the full texts of potentially relevant studies against the inclusion criteria and any disagreements resolved as above.

2.1.6 Quality appraisal

I subjected each of the included studies to quality appraisal using the Critical Appraisal Skills Programme (CASP, 2018). The CASP tool comprises 10 questions (shown in Appendix 3) and is suitable for use with any methodology. The CASP does not include a scoring system and so one was not used. A scoring system has been suggested (Butler et al., 2016), but arbitrary quantification of quality and the creation of cut-off scores for inclusion have been deemed unnecessary and may lead to false conclusions and unwarranted exclusions, partly due to the importance of quality being unequal across domains (Noyes et al., 2019). Others do choose to exclude studies on the basis of quality (Tong et al., 2012, Carroll et al., 2012, Noyes and Popay, 2007, Toye et al., 2013), though thresholds are specific to individual reviews.

There appeared to be little guidance on the application of CASP, also recognised by Long et al. (2020), who identified further limitations in the tools ability to assess the philosophical positions of researchers within individual studies. Long et al. (2020) went on to modify the tool with an additional question to allow for greater clarity in assessing the philosophical position of studies and how this translated to their methods and methodologies. Furthermore, Long et al. (2020) added an additional response to the existing “yes”, “no” and “can’t tell”. The addition of “somewhat” allowed a more nuanced approach to answering the questions where “somewhat” meant there had been a reasonable attempt at fulfilling the criteria and “can’t tell” meant that there was not enough information to make a judgment. Long et al. (2020) also described a process by which they coded studies in an order based upon quality. Studies deemed to be of high quality were coded first, followed by those of medium quality and finally those of low quality. They used their existing coding framework to code the latter studies, meaning that no additional codes were added for low quality studies and they had less weighting in the overall synthesis. Weighting according to quality has been recognised as allowing appraisals to make a meaningful contribution to the overall synthesis (Boeije et al., 2011). Similarly, post-hoc sensitivity analyses can help authors to identify studies’ contributions to syntheses by judging how their exclusion affects results. Several studies have found that lower quality studies make minimal contribution to final results (Carroll et al., 2012).

I coded studies according to diagnosis, rather than quality, though the CASP was used to enable me to engage with the strengths and limitations of the included studies, their credibility and weighting of their conclusions and how this may have impacted upon the overall synthesis. A full sensitivity analysis was not carried out, but I made an assessment as to the coding contributions of the lower quality studies, whether they confirmed the findings of the higher quality studies and whether or not they provided any new or unique insights. Critical appraisal was carried out by myself and a second reviewer assessed 30% of the studies to determine agreement.

2.1.7 Data extraction

I used a data extraction table to capture key characteristics of each study, including authors, year of publication, year of data collection, country, setting, aims, methodology and methods and sample characteristics. I then extracted all data labelled as ‘findings’, ‘results’ or pertaining to such, such as results reported in the discussion, to NVivo. This included authors’ interpretations as well as quotes from participants. Several approaches to data extraction have been suggested and range from inclusive approaches, in which all eligible data are included, to a more selective approach in which data is extracted on the basis of theoretical relevance or validity as considered by the reviewer. The inclusive approach taken here was appropriate given the aim of gaining a deeper understanding of the experiences of fathers in the absence of an *a priori* hypothesis and an approach consistent with thematic synthesis (Thomas and Harden, 2008).

2.1.8 Conducting the synthesis

I followed the stages of thematic synthesis (Thomas and Harden, 2008) in which text is coded line-by-line to create a bank of codes and concepts that can be translated across studies. More than one code can be assigned to a line and new codes should be added when necessary. I developed descriptive themes by grouping codes together based on similarities and used new codes to define each group and to form a hierarchical structure. I then developed analytical themes to "generate new interpretive constructs, explanations or hypotheses" (Thomas and Harden, 2008). I carried out coding with regular input from the review team during the development of descriptive and analytical themes.

2.2 Results

2.2.1 Study selection results

Figure 2 shows a Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) diagram of the search results and reasons for exclusion. The electronic database searches identified 4273 unique results. I removed 4212 of these studies during title and abstract screening. I screened the full texts of the remaining 63 papers against the eligibility

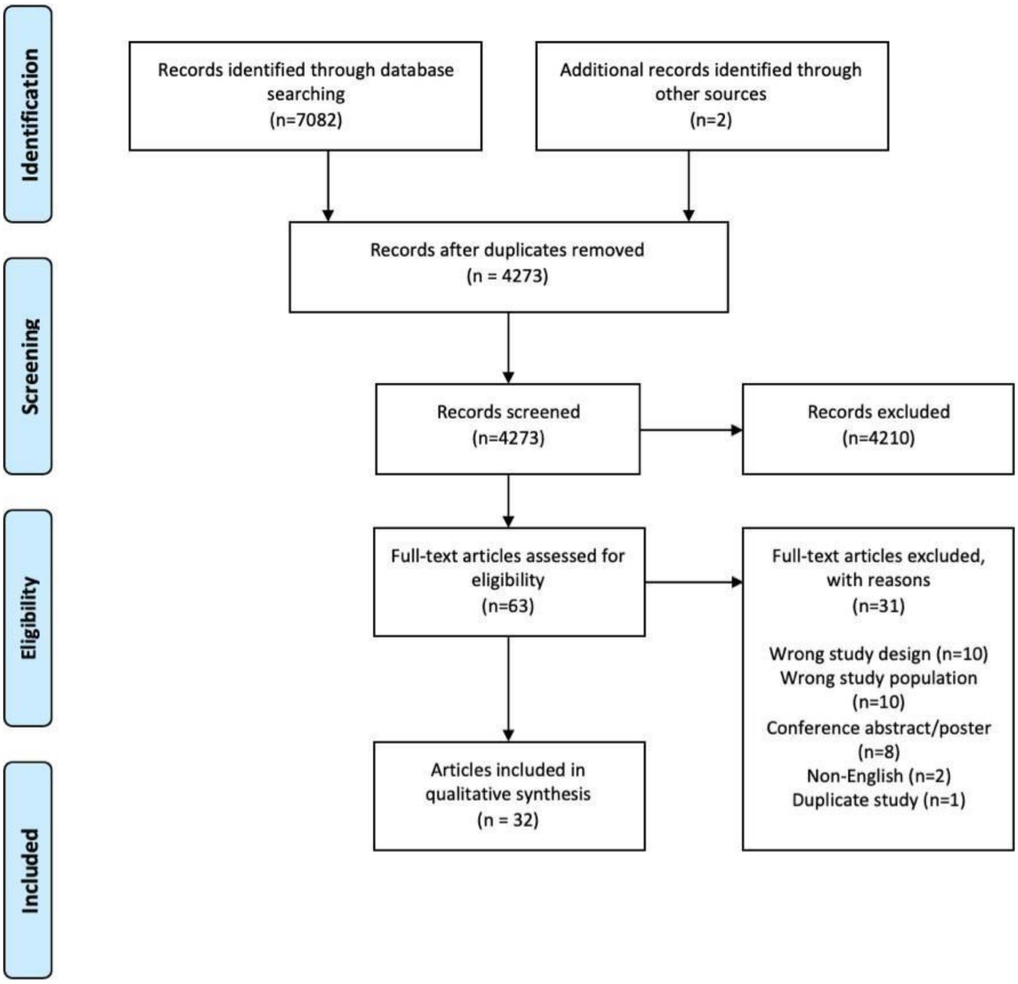


Figure 2; PRISMA flowchart showing the inclusion of 32 articles from 30 studies criteria from which a further 31 papers were excluded. The final thematic synthesis consisted of 32 eligible papers from 30 studies.

2.2.2 Quality appraisal

Most of the studies included in the review were of medium (12) to high (16) quality. Four studies were deemed to be of low quality. These 'high', 'medium' and 'low' ratings were ascribed to the included studies, despite not using a scoring system, and were based upon the overall answers to questions in the CASP tool. They were intended to improve readability and to summarise the quality appraisal findings. These ratings were discussed and agreed on with the second reviewer. The quality appraisal table can be seen in Appendix 3.

2.2.3 Study characteristics

The qualitative studies included in the review were published between 1997 and 2019 with data collection taking place between 1978 and 2019. The majority of the included papers were published in the US (18), followed by the UK (4), Canada (3) and Australia (2). There was 1 paper each from New Zealand, Ireland, Hong Kong, Sweden and Brazil. 27 studies were published in peer review journals, 5 were PhD theses. See table 3 for details of study characteristics.

Eight studies focused on fathers' lived experiences and fatherhood in the context of having a child with a life-limiting condition (Appelbaum and Smolowitz, 2012, Gower et al., 2016, Lucca and Petean, 2016, Nicholas et al., 2009, Nicholas et al., 2016, Neil-Urban and Jones, 2002, Jones and Neil-Urban, 2003, Rivard and Mastel-Smith, 2014). Four focused on resilience and coping (Brody and Simmons, 2007, Chamberlain, 2007, Wills, 2009, Wolff et al., 2010) and three studies examined fathers' experiences and perceptions of service provision and support (Nicholas et al., 2016, Bruce et al., 2016, Ware and Raval, 2007). One study examined the influence of gender on fathers' experiences (Chesler and Parry, 2001) and one examined their experiences through an attachment lens (Bailey-Pearce et al., 2017). Three studies focused on fathers' experiences around the time of diagnosis (Clark and Miles, 1999, Ogg, 1997, Priddis et al., 2010). Seven studies focused on fathers' caregiving experiences or their perception of their role (Cluley, 2015, Hill et al., 2009, Robinson et al., 2019b, Shardonofsky et al., 2019, Clarke, 2005, Wolff et al., 2011, Mojica, 2016, Robinson et al., 2019a) and one the emotional impact of managing care (Hayes and Savage, 2008). Three studies explored the experiences of bereaved father's (Rigby, 2012, Davies et al., 2004, Davies et al., 2013) (see table 5).

2.2.4 Participant & child characteristics

15 papers (from 13 studies) concerned the experiences of fathers of children with cancer (Brody and Simmons, 2007, Chesler and Parry, 2001, Clarke, 2005, Cluley, 2015, Mojica, 2016, Jones and Neil-Urban, 2003, Neil-Urban and Jones, 2002, Ogg, 1997, Robinson et al., 2019b, Wills, 2009, Wolff et al., 2010, Chamberlain, 2007, Hill et al., 2009, Nicholas et al., 2009, Wolff et al., 2011), five concerned the experiences of fathers of children with a congenital heart defect (Bright et al., 2013, Bruce et al., 2016, Clark and Miles, 1999, Gower et al., 2016, Robinson et al., 2019a), three concerned fathers of children with cystic fibrosis (Hayes and Savage, 2008, Priddis et al., 2010, Shardonofsky et al., 2019), two focused on fathers of children with neurological conditions (Appelbaum and Smolowitz, 2012, Lucca and Petean, 2016) and one study concerned the experiences of fathers with a genetic condition (Rivard and Mastel-Smith, 2014). Six studies were not diagnosis specific and included fathers of children with a range of life-limiting and life-threatening conditions including, but not limited to, cancer, cystic fibrosis and spinal muscular atrophy (Bailey-Pearce et al., 2017, Davies et al., 2004, Nicholas et al., 2016, Ware and Raval, 2007, Davies et al., 2013, Rigby, 2012). The children were aged between infancy and 27 years but were all diagnosed in childhood.

The included papers represented the experiences of 562 fathers including biological fathers, step-fathers, adoptive fathers. This included bereaved fathers. Sample sizes for each individual paper can be seen in table 3 and range from 6-167. From the demographics that were reported the age range of the fathers was 23-65 years. They came from a range of educational backgrounds and occupations. The majority were married/cohabiting with the mother of their child/ren and were in employment.

Table 3; study characteristics of the included studies

Study	Aim	Year of data collection	Country	Setting	Methodology	Data collection	Data analysis
Cancer							
Brody and Simmons (2007)	To explore the challenges that fathers face following their child's cancer diagnosis, how they cope with and adapt to this diagnosis.	Not stated	US	University pediatric oncology clinic, Kentucky	Qualitative descriptive design	Semi-structured interviews	Open coding
Chamberlain (2007)	To explore resilience in fathers of children with leukemia	Not stated	US	Leukemia & Lymphoma society	Not stated	Semi-structured interviews	Grounded theory
Chesler and Parry (2001)	To explore the experiences of fathers of children with cancer with a particular focus on how such	1978-1998	US	Participants identified from survey data (1978-1998), focus	Not stated	In-depth interviews, workshops	Deductive coding using stress framework followed by

Study	Aim	Year of data collection	Country	Setting	Methodology	Data collection	Data analysis
	experiences are influenced by gender.			group conducted at conference			inductive coding using a grounded theory approach
Clarke (2005)	To describe fathers' experiences of having a child with cancer, with a focus on home healthcare work concerns.	Not stated	Canada	Recruited through support groups/ partners/ researchers personal network. Child treat in geographical location in past 5 years	Not stated	Qualitative interviews	Deductive coding based on home healthcare work

Study	Aim	Year of data collection	Country	Setting	Methodology	Data collection	Data analysis
Cluley (2015)	To explore the experiences of fathers of children with cancer and how they understand their role in their child's illness	Not stated	New Zealand	Child Cancer Foundation	Narrative inquiry-social constructionist approach	Narrative interviews	Narrative analysis
Hill et al. (2009)	To explore fathers' perceptions of their role during their child's treatment for acute lymphoblastic lymphoma	Not stated	Northern Ireland	Treating hospital	Phenomenology	Semi-structured interviews	IPA
Jones and Neil-Urban (2003)	To investigate the experiences of fathers of children with cancer with a focus on the caregiving experience	Not stated	US	Hospital	Group phenomenological approach	Focus groups	Grounded theory, constant comparison method, group process analysis (secondary analysis)

Study	Aim	Year of data collection	Country	Setting	Methodology	Data collection	Data analysis
Mojica (2016)	To explore masculinity, caregiving and coping in the context of having a child with cancer	Not stated	US	Paediatric hospital	Mixed methods	Open-ended questions	Content analysis
Neil-Urban and Jones (2002)	To describe the experiences and coping of fathers who have a child with cancer	Not stated	US	Hospital	Group phenomenological approach	Focus groups	Thematic approach (Van Maanen, 1983)
Nicholas et al. (2009)	To examine experiences of fatherhood in the context of childhood cancer from the perspectives of fathers	Not stated	Canada	Central pediatric hospital	Grounded Theory	Semi-structured interviews	Grounded Theory
Ogg (1997)	To examine the effect of a paediatric cancer diagnosis from fathers' perspectives	Not stated	US	Hematology/ oncology outpatient clinic at medical center	Not stated	Structured interviews	Grounded Theory

Study	Aim	Year of data collection	Country	Setting	Methodology	Data collection	Data analysis
Robinson et al. (2019b)	To explore the paternal roles, responsibilities, strengths, challenges, personal growth and support needs of fathers of children with brain tumours.	December 2018- February 2019	US	Pediatric Hospital	Not stated	Semi-structured interviews	Semantic content analysis
Wolff et al. (2010)	To look at how fathers describe their experiences and challenges, sources of support and coping and unique challenges described by single fathers and those from racial and ethnic minority groups.	Not stated	US	Pediatric Hospital	Life-story method	Semi-structured interviews	Narrative analysis with multiple case studies

Study	Aim	Year of data collection	Country	Setting	Methodology	Data collection	Data analysis
Wolff et al. (2011)	To understand why fathers assume the primary caregiving role to their child with a life-threatening illness; to examine how they describe their reasons for assuming this role and how race, ethnicity and socioeconomic status shape this decision	Not stated	US	Pediatric Hospital	Life-story method	Semi-structured interviews	Narrative analysis with multiple case studies
Wills (2009)	To describe the experiences coping strategies of fathers of children with acute lymphocytic leukemia	Not stated	China	Pediatric oncology ward, large teaching hospital, Hong Kong	Not stated	Semi-structured interviews	Qualitative data analysis using matrix system (Miles & Hubermann, 1994)
Congenital Heart Defect							

Study	Aim	Year of data collection	Country	Setting	Methodology	Data collection	Data analysis
Bright et al. (2013)	To examine the relationship between fathers and their infant with CHD	Not stated	Australia	Paediatric hospital	Mixed methods	Interviews	Qualitative analysis
Bruce et al. (2016)	To explore the experiences and meaning of support from the perspectives of fathers of children with congenital heart defects	2009	Sweden	Paediatric cardiac outpatient clinic	Phenomenological-hermeneutic	Narrative interviews	Phenomenological-hermeneutic method for interpreting narrative interviews (Lindseth &Norberg (2004))
Clark and Miles (1999)	To explore the experiences of fathers whose infants were diagnosed with	Not stated	US	NICU, ICU, pediatric ICU	Not stated	Longitudinal semi-structured interviews at	Content analysis

Study	Aim	Year of data collection	Country	Setting	Methodology	Data collection	Data analysis
	severe congenital heart disease					diagnosis and 12 months post-diagnosis	
Gower et al. (2016)	To explore the lived experiences of fathers of children with a congenital heart defect	Not stated	UK	Regional clinic	Not stated	Semi-structured interviews	IPA
Robinson (2019)	To understand the needs, role understanding and resiliency in fathers of children receiving cardiac care with a new palliative care referral	Not stated	US	Children's hospital	Not stated	Interviews	Semantic content analysis
Cystic fibrosis							
Hayes and Savage	(2008)	To examine the		emotional impact of	caring for a child	Not stated	I reland

Cystic fibrosis center	Not stated	Qualitative interviews	Thematic content analysis (Burnard, 1991)

Study	Aim	Year of data collection	Country	Setting	Methodology	Data collection	Data analysis
	with CF from fathers' perspectives						
Priddis et al. (2010)	To explore fathers' perceptions of the familial impact of their infant's CF diagnosis	Not stated	Australia	Paediatric care provider	Not stated	Semi-structured interviews	Qualitative content analysis (Smith, 2001)
Shardonofsky et al. (2019)	To explore fathers' perspectives of caring for a child with cystic fibrosis	Not stated	US	Cystic fibrosis centres	Descriptive Phenomenology (Husserl, 1970)	Semi-structured interviews	Descriptive phenomenology (Giorgi, 1985)
Genetic conditions							
Rivard and Mastel-Smith (2014)	To describe the experiences of fathers who have children diagnosed with a genetic disorder.	Recruitment April-October 2012	US	Identified via nurses, physicians, genetic counsellors, snowball sampling,	Phenomenology	Semi-structured interviews	IPA

Study	Aim	Year of data collection	Country	Setting	Methodology	Data collection	Data analysis
				email, internet, word-of-mouth			
Life-limiting conditions							
Bailey-Pearce et al. (2017)	To explore the experiences of fathers of children with a LLC and to examine how their attachment strategies influence these experiences.	Not stated	UK	Identified via community paediatric nursing team	Narrative approach	Narrative interviews	Narrative analysis using Riessman's methodology (2008)
Davies et al. (2013)	To explore fathers' perspectives of the care received from healthcare professionals during their child's illness and death	Not stated	US	Paediatric palliative care settings	Grounded Theory	Semi-structured interviews, field notes, genograms, reflexive journaling,	Open coding, constant comparative analysis

Study	Aim	Year of data collection	Country	Setting	Methodology	Data collection	Data analysis
						interview debriefing	
Davies et al. (2004)	To provide a description and theoretical explanation of fathers' experiences of their child's life-limiting illness	Not stated	US	Pediatric hospice home care program	Grounded Theory	In-depth unstructured interviews	Grounded Theory
Nicholas et al. (2016)	To explore father's experience and support provision preferences in relation to their child's life-limiting illness	Not stated	Canada	Tertiary level paediatric hospitals	Grounded Theory	Semi-structured interviews	Grounded Theory
Rigby (2012)	To explore the experiences and grief of fathers following the loss of their child to a life-limiting condition.	Not stated	US	Recruitment across 3 states	Secondary analysis	Semi-structured, in-depth and follow-up interviews	Qualitative techniques (Dey, 1993)

Study	Aim	Year of data collection	Country	Setting	Methodology	Data collection	Data analysis
Ware and Raval (2007)	To investigate the experiences of fathers of children with a life-limiting illness and how it has affected them and their family relationships. To explore their perceptions of service provision and how/if services could be made more readily available to them.	Not stated	UK	Recruited via several sources (newsletter, parent support groups, voluntary organisations)	Phenomenology	Semi-structured interviews	IPA (Smith, 1995)
Neurological conditions							
Appelbaum and Smolowitz (2012)	To examine the experiences of fathers of children with severe cerebral palsy.	Not stated	US	Referred via healthcare staff at residential facility	Phenomenology	Interviews	Thematic analysis using van Maanen's methodology (1990)

Study	Aim	Year of data collection	Country	Setting	Methodology	Data collection	Data analysis
Lucca and Petean (2016)	To understand the experiences of fathers of children with Duchenne Muscular Dystrophy (DMD)	Not stated	Brazil	Association of Muscular Dystrophy in Ribeirão Preto	Not stated	Semi-structured interviews	Content analysis

2.2.5 Thematic synthesis

Line-by-line coding produced 245 codes. I identified similarities between the codes and grouped them into 8 descriptive themes (table 6a). These themes captured fathers' experiences of having a child with a life-limiting condition including their response to diagnosis, impact on day-to-day life, workplace factors, emotional wellbeing, relationships with family and friends, relationships with professionals, their experiences and preferences for support, and thinking about the future and their child's death. Through the synthesis, I organised the data into 4 analytical themes (table 6b): (1) uncertainty; introduction and adaptation; (2) professionals reinforcing fathers' role perceptions; (3) working fathers; role conflict; (4) a new normal.

Tables 4a and 4b; descriptive and analytical themes

Descriptive themes
Diagnosis
Day-to-day-life
Workplace support
Emotional experiences
Relationship with professionals
Relationship with family and friends
The future
Death

Analytical themes
Uncertainty; introduction and adaptation
Professionals reinforcing fathers' role perceptions
Working fathers; role conflict
A new normal

2.2.5.1 *Theme 1: Uncertainty; introduction and adaptation*

A key theme describes the uncertainty that dominated fathers' accounts of their child's condition. Three sub-themes (figure 2), organised temporally, make up the theme, and describe the introduction of a new uncertainty pre-diagnosis, a transition to a new form of uncertainty *at* diagnosis and finally adaptation to this uncertainty.

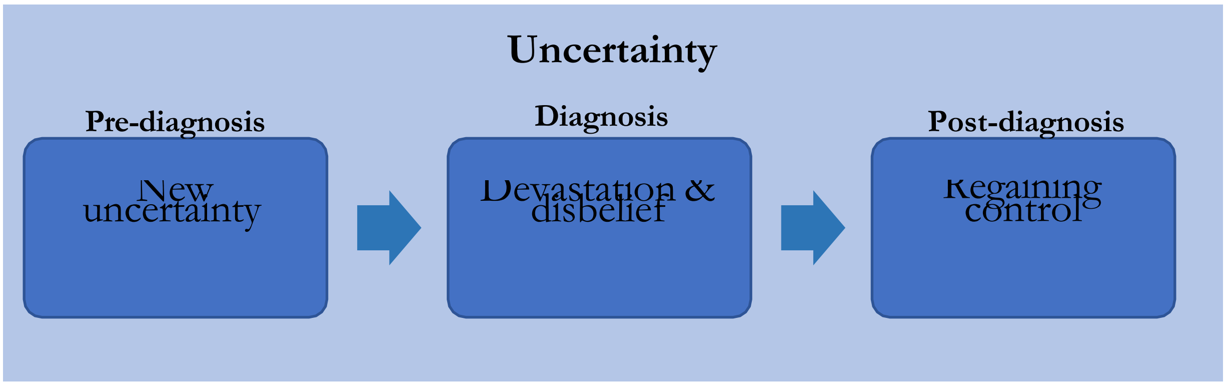


Figure 3; Theme 1- uncertainty: introduction and adaptation

2.2.5.1.1 New uncertainty

The period leading up to diagnosis varied for fathers, though many endure difficult “battles”, multiple hospital visits, demanding conversations and long waits in order to get a confirmed diagnosis (Ware and Raval, 2007, Clarke, 2005, Jones and Neil-Urban, 2003, Brody and Simmons, 2007, Lucca and Petean, 2016, Bailey-Pearce et al., 2017). With this waiting came frustration, sometimes directed towards professionals if they did not appear to be providing fathers with adequate information.

“Delay in diagnosis was due to several causes: inadequate medical resources; current state of knowledge of childhood cancers; and, in one case, turf issues between medical personnel” (Jones and Neil-Urban, 2003) (cancer, US).

“There were moments of uncertainty while their child either presented with an unusual health issue or were born with an immediate medical need and needed intensive care; this uncertainty continued as their child went through tests” (Bailey-Pearce et al., 2017) (life-limiting condition, UK).

“In the days and weeks leading up to the diagnosis, a few fathers indicated having to take their child to the doctor numerous times before a diagnosis was made. These instances were challenging to fathers because it was frustrating that the doctors could not immediately tell them why their child was so sick” (Brody and Simmons, 2007) (cancer, US).

This new uncertainty often left fathers with feelings of helplessness (Hill et al., 2009, Bailey-Pearce et al., 2017, Lucca and Petean, 2016, Cluley, 2015, Chamberlain, 2007, Nicholas et al., 2009). This sense of helplessness pervaded fathers accounts, not only in relation to having wait for a potentially devastating diagnosis, but in wanting to protect and help their child.

The "delay" and long time that passed between the onset of symptoms and diagnosis, combined with various exams the child has to take, generate anxiety, anguish, and uncertainty (Lucca and Petean, 2016) (Duchenne Muscular Dystrophy, Brazil).

"The bump was discussed I would say approximately in the middle of June. It seemed to take longer than we felt was appropriate. We got the biopsy results in August. Even with my friend John (a doctor), he kind of pulled strings and pushed people along, it still seemed to take a long time. It was pretty troubling waiting that amount of time. That was probably the hardest part, waiting" (Clarke, 2005) (cancer, Canada).

2.2.5.1.2 Diagnosis; devastation & disbelief

Having a child diagnosed with a life-limiting condition was shocking (Clarke, 2005, Appelbaum and Smolowitz, 2012, Ware and Raval, 2007, Rivard and Mastel-Smith, 2014, Jones and Neil-Urban, 2003, Lucca and Petean, 2016, Wills, 2009, Bailey-Pearce et al., 2017, Brody and Simmons, 2007, Chamberlain, 2007). As discussed above, the wait for a diagnosis was often difficult and lengthy, with some fathers "battling" for answers (Clarke, 2005, Lucca and Petean, 2016, Neil-Urban and Jones, 2002, Jones and Neil-Urban, 2003, Rivard and Mastel-Smith, 2014, Brody and Simmons, 2007).

Despite some diagnoses confirming fathers' fears, this sense of shock was prominent in their accounts, perhaps demonstrating a sense of hope pre-diagnosis. Many fathers were unable to comprehend the reality of what had happened to their child (Priddis et al., 2010, Wills, 2009, Davies et al., 2004, Appelbaum and Smolowitz, 2012, Cluley, 2015, Ware and Raval, 2007, Bailey-Pearce et al., 2017, Hill et al., 2009). There was a preconception that "things like this don't happen to my family" (Wills, 2009). The helplessness that some fathers experienced as they awaited their child's diagnosis was sustained post-diagnosis (Clark and Miles, 1999, Clarke, 2005, Cluley, 2015, Davies et al., 2004, Lucca and Petean, 2016, Nicholas et al., 2009, Hill et al., 2009).

“You feel that you’ve been taken out of your life and put into somebody else’s movie… the wrong movie. Like if you were watching a film in the theatre and when they change the reels, they put on a reel from a different movie ... it’s that disorienting. It’s a very alienating experience” (Davies et al., 2004) (life-limiting conditions, US).

In the beginning, I was just a young man who was going to the hospital with my wife to have a baby. It was something I didn’t expect was going happen to me and my family (Appelbaum and Smolowitz, 2012) (cerebral palsy, US).

Fathers described diagnosis as a traumatic event; like being “thrown into a hurricane” (Rigby, 2012), like a bomb (Lucca and Petean, 2016), and like being “smacked in the guts with a sledgehammer” (Cluley, 2015). These vivid accounts (Bailey-Pearce et al., 2017, Ware and Raval, 2007), were contrasted by fairly vague descriptions of diagnosis, blurred by shock or concern over their partners emotional response (Chamberlain, 2007, Lucca and Petean, 2016, Priddis et al., 2010).

Fathers expressed a range of negative emotions in response to their child’s diagnosis such as **sadness** (Clark and Miles, 1999), **anger** (Bailey-Pearce et al., 2017, Rivard and Mastel-Smith, 2014), and **devastation** (Jones and Neil-Urban, 2003, Bailey-Pearce et al., 2017, Cluley, 2015, Priddis et al., 2010, Ware and Raval, 2007). Some described being overwhelmed by events (Shardonofsky et al., 2019, Ware and Raval, 2007) and some described feelings of grief and loss over their child’s future, comparing their child to others (Appelbaum and Smolowitz, 2012, Chamberlain, 2007, Jones and Neil-Urban, 2003, Lucca and Petean, 2016, Rigby, 2012, Ware and Raval, 2007), and loss of their expected experience of fatherhood (Ware and Raval, 2007).

‘A feeling of devastation, yes, you know that somehow the world changed from what it was an hour and a half before’ (Ware and Raval, 2007) (life-limiting condition, UK).

"I think you always do have regrets when there are other children of my friends and family that are my daughter's age. They go to a prom or they're driving the first time. I think about those things, sometimes. But I feel bad because she won't be able to experience being a mother" (Appelbaum and Smolowitz, 2012) (cerebral palsy, US).

2.2.5.1.3 Regaining control

The uncertainty that fathers experienced in the period leading up to their child's diagnosis was not resolved once they received the diagnosis. It was transformed into uncertainties relating to their child's condition and treatment, including potential causes of illness, and the future. Fathers struggled to make sense of their child's diagnosis and information seeking played a large part in their behavioural response to diagnosis (Jones and Neil-Urban, 2003). For some, there was an initial transient form of information seeking, that was in relation to finding an explanation or seeking to attribute blame to something or someone for their child's condition (Wills, 2009, Cluley, 2015, Davies et al., 2004, Jones and Neil-Urban, 2003, Rivard and Mastel-Smith, 2014).

''We had a dog, did the kid play with the dog too much?!'' Some wondered if they themselves had done something wrong: ''I did construction on the house. Did I bring some contaminated material into the house?'' Or, they wondered if their genes were faulty in some way. For fathers of children with genetic disease, this was an especially poignant reality. However, for some it was a great relief to know that the illness was genetic as it meant that there was nothing they could have done to prevent it" (Davies et al., 2004) (life-limiting conditions, US). *"I blamed myself because I should have told him not to eat cup noodles for lunch every day. The preservatives might have caused his disease"* (Wills, 2009) (cancer, Hong Kong).

Upon further examination of the data, it became clear that this explanation/blame seeking was soon replaced with seeking information regarding prognosis, trajectory and living with the condition which could feel more productive or useful for fathers. Part of this information seeking helped fathers to assert some control in areas that they felt could be managed. Fathers straightforwardly described focusing on "what needed to be done" (Brody and Simmons, 2007), the "here and now" (Priddis et al., 2010), "tackling things head on" (Nicholas et al., 2016), "getting on with it" (Hill et al., 2009) and a determination that the illness could be "beaten" (Rigby, 2012). Fathers sought to regain control through understanding and began to consider that uncertainty would be a part of their lives, and despite the unsettling nature of this uncertainty, accepting and adapting.

“[The] lingering uncertainty means that I’ll never totally bounce back. The uncertainty of not knowing what’s going to happen, the fear he’s going to die. But most challenging is the uncertainty. It’ll be with me indefinitely” (Chamberlain, 2007) (cancer, US).

“… in the last few years I've found I became more positive you know, trying to… not live with kind of a cure, just trying to live with it… the bottom line is… none of us know what is going to happen to us tomorrow anyhow” (Hayes and Savage, 2008) (cystic fibrosis, Ireland).

“One way in which the fathers dealt with the unpredictability of the disease and their lack of control over it was to set their sights on more immediate and achievable goals, such as becoming the advocate of their child” (Jones and Neil-Urban, 2003) (cancer, US).

There was a spectrum of informational need from fathers, with some seeking to understand all that they could about their child’s condition through a great deal of self-directed research and some seeking only the information that would allow them to address current challenges. Both strategies seemed to originate from the fear of the unknown; some wanted as little ‘unknown’ as possible whilst some did not want to know about things that could potentially happen in the future for which they had no control over.

“The fathers spoke of putting their energies into making the best of the time they had together as a family and not worrying about what could happen” (Hayes and Savage, 2008) (cystic fibrosis, Ireland).

“Another coping strategy was to concentrate primarily on short-term goals, such as planning your child’s next birthday party and focusing on the most recent treatment success rather than on the long range prospects” (Jones and Neil-Urban, 2003) (cancer, US).

2.2.5.2 *Theme 2: Professionals; reinforcing fathers' role perceptions*

In this second theme, I incorporated two concepts; how fathers perceived their role and emotional needs, and how such perceptions were often reinforced through their interactions with healthcare professionals. Findings are first presented in relation to each subordinate theme which are then integrated to demonstrate how the lens through which fathers saw their role was reinforced by professionals. The relationship between these subordinate concepts feeds into the 'strong, silent and supportive' narrative seen across fathers' accounts.

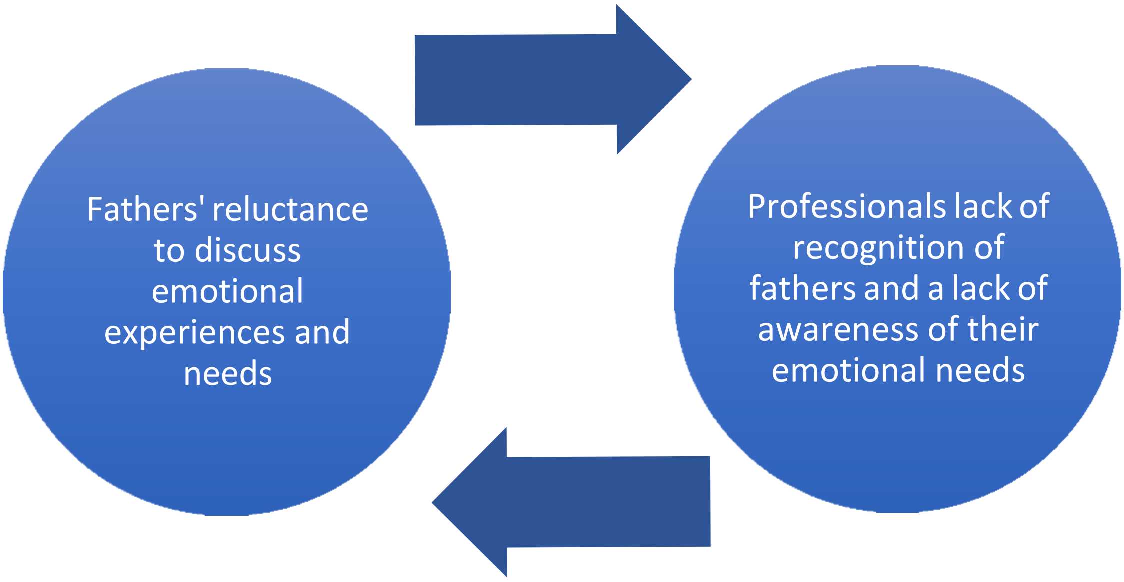


Figure 4; Theme 2: reciprocal relationship between fathers' feelings and experiences/interactions with HCP's

2.2.5.2.1 Keeping emotions hidden from others

Fathers experienced a range of emotions and challenges in their everyday lives at, and beyond, diagnosis. Fathers discussed the overwhelming and turbulent nature of their emotional worlds (Brody and Simmons, 2007, Nicholas et al., 2016, Bailey-Pearce et al., 2017, Chesler and Parry, 2001). They described mental strain (Brody and Simmons, 2007), exhaustion (Chamberlain, 2007, Bruce et al., 2016, Clarke, 2005, Gower et al., 2016, Rivard and Mastel-Smith, 2014, Nicholas et al., 2009), loneliness (Chamberlain, 2007, Chesler and Parry, 2001, Nicholas et al., 2009, Nicholas et al., 2016, Shardonofsky et al., 2019, Bruce et al., 2016, Jones and Neil-Urban, 2003, Ware and Raval, 2007), vulnerability (Bruce et al., 2016, Davies et al., 2004) and sadness (Bruce et al., 2016, Nicholas et al., 2016, Chesler and Parry, 2001, Clarke, 2005).

"I try to keep my cool because you will just, it's not that you mean to do it, you just snap because it's too much, you can't handle it, it's too much" (Nicholas et al., 2016) (life-limiting conditions, Canada).

"Liam described how he is affected emotionally by the illness spontaneously and acknowledges how his emotional world affects his thoughts about the future: I get really upset about it sometimes often at the most random time yeh you'd be having a completely normal time and then all of a sudden it hits you oh my god" (Bailey-Pearce et al., 2017) (life-limiting conditions, UK).

Fathers also described an underlying feeling of anxiety related to a fear of bad news (Clark and Miles, 1999, Hayes and Savage, 2008, Nicholas et al., 2016, Chesler and Parry, 2001), the trajectory of the illness (Gower et al., 2016, Hayes and Savage, 2008, Rivard and Mastel-Smith, 2014), and relapse (Chamberlain, 2007). Some fathers also described the constant and underlying fear they had of exposing their child to unnecessary risk or missing important symptoms, resulting in a state of hyper-vigilance and further contributing to their exhaustion (Chamberlain, 2007, Jones and Neil-Urban, 2003, Hayes and Savage, 2008, Gower et al., 2016, Hill et al., 2009, Bailey-Pearce et al., 2017, Brody and Simmons, 2007, Clarke, 2005, Neil-Urban and Jones, 2002).

"One father stated, "Chances are, that with that syndrome, they'll never be on their own, they can't be." Another commented, "You're so tired. I mean, we still wake up every night with [our child], you feel like a zombie"" (Rivard and Mastel-Smith, 2014) (Genetic conditions, US).

"There was a sense of exhaustion from constant hyper-vigilance while taking responsibility for their child's safety and being the omnipotent protector proved a tough task, from which it was difficult to escape" (Gower et al., 2016) (congenital heart defects, UK).

Fathers discussed keeping these emotional struggles hidden from their families. For some, this was due to a belief that they needed to be strong and supportive of their partners' and children, who were the priority, and that in exhibiting their own distress, they would cause further suffering to those around them (Chamberlain, 2007, Bailey-Pearce et al., 2017, Chesler and Parry, 2001, Clark and Miles, 1999, Gower et al., 2016, Hayes and Savage, 2008, Hill et al., 2009, Lucca and Petean, 2016, Nicholas et al., 2009, Priddis et al., 2010, Rigby, 2012, Ware and Raval, 2007, Nicholas et al., 2016).

For some, this was more explicitly tied to fathers' perceptions of masculinity. They believed that it was not part of a man's role as protector to express emotion, with some associating it with weakness and femininity, focusing on the needs of others as to deflect from their own experiences (Bailey-Pearce et al., 2017, Hayes and Savage, 2008, Hill et al., 2009, Ware and Raval, 2007, Gower et al., 2016, Nicholas et al., 2009, Jones and Neil-Urban, 2003, Lucca and Petean, 2016, Neil-Urban and Jones, 2002).

"I said to Gillian [mother] 'right I need to go out and I'll phone your Mum and phone my mum and tell them' and again that was me I had to get away from her because I wanted to cry" (Hill et al., 2009) (cancer, UK).

"In general, fathers felt it was their role within the family to maintain equilibrium and strength, and in doing so, they sought to support their partner, which included suppressing their own emotions and negative feelings" (Nicholas et al., 2009) (cancer, Canada).

"It may have also served as protection from potentially overwhelming emotions which could jeopardise the role of container; perhaps a focus on practicalities provided an escape from their own emotional responses so they could avoid upsetting others" (Gower et al., 2016) (congenital heart defect, UK).

Others further described this emotional suppression as a means of minimising their own suffering; to discuss their distress would be too difficult and they did not have the tools to do so (Chesler and Parry, 2001, Nicholas et al., 2009, Lucca and Petean, 2016, Bailey-Pearce et al., 2017, Hayes and Savage, 2008).

This was in contrast with some fathers' perceptions that emotional expression was acceptable (Hill et al., 2009) and experiences when doing so had been helpful (Hill et al., 2009, Chamberlain, 2007). Some fathers felt that, in retrospect, emotional support would have been beneficial during more difficult times (Priddis et al., 2010). Furthermore, some fathers expressed a current desire to discuss how they were feeling but felt that they didn't have an appropriate confidant or didn't know where to seek more formal support.

2.2.5.2.2 Fathers; the forgotten parent in healthcare settings

This perception of fathers being a supportive figure for others was reinforced in healthcare settings. Fathers discussed their interactions with professionals with respect to what they had experienced more generally as a family, as well as more specifically to experiences that they had perceived as being unique to them as fathers. Some described a lack of recognition from professionals and felt that healthcare systems were predominantly biased towards engaging with mothers, leaving fathers on the periphery of their child's care (Bailey-Pearce et al., 2017, Brody and Simmons, 2007, Chesler and Parry, 2001, Gower et al., 2016, Hill et al., 2009, Nicholas et al., 2016, Davies et al., 2013, Ware and Raval, 2007, Appelbaum and Smolowitz, 2012, Rivard and Mastel-Smith, 2014, Priddis et al., 2010, Nicholas et al., 2009, Clarke, 2005). This lack of interaction and communication resulted in fathers feeling helpless, surplus to requirement, isolated and out-of-control (Brody and Simmons, 2007, Nicholas et al., 2016, Ware and Raval, 2007, Gower et al., 2016, Rivard and Mastel-Smith, 2014).

"As a result, many fathers did not receive needed support and felt left out of the direct chain of medical information" (Chesler and Parry, 2001) (cancer, US).

"Fathers experienced intense emotion; however, they described themselves as "alone," "strong," and "to themselves"" (Nicholas et al., 2009) (cancer, Canada).

In contrast to this, feeling involved and understood by professionals seemed to help fathers to cope with the situation they faced; regardless of the news they received, if information was clear, they felt a greater sense of control. The opposite was demonstrated in the uncertainty and fear that they experienced when professionals did not communicate effectively or did not involve fathers in decision making (Bruce et al., 2016, Clarke, 2005, Davies et al., 2004, Gower et al., 2016). This emphasized the importance of fathers feeling listened to, understood and an integral part of their child's care rather than an "observer" (Gower et al., 2016) or "spectator" (Rivard and Mastel-Smith, 2014). Fathers value straightforward, honest, knowledgeable, approachable and calm professionals (Jones and Neil-Urban, 2003, Bruce et al., 2016, Brody and Simmons, 2007, Davies et al., 2004).

". . . when I got the bad news . . . it was like a stone was falling on me . . . yes . . . it was heavy—heavy—but at the same time, it was great to have a picture of what was wrong [clears his throat]" (Bruce et al., 2016) (congenital heart defect, Sweden).

"Fathers also feel safe and recognized as an important member of the family when health care professionals not only give their time but also stand by the fathers' side and help them fight for their families' right to get information and be together" (Bruce et al., 2016)

2.2.5.2.3 Integration of sub-themes

Fathers described this need for effective communication, and in the absence of such felt a great sense of loneliness and like an observer in the care of their child. In prioritizing the needs of their family through this 'strong and silent' mindset, they set themselves at the periphery of care as a supportive figure, and their own needs and opinions were overlooked, both in relation to their child's care and their own wellbeing (Chesler and Parry, 2001, Hayes and Savage, 2008, Hill et al., 2009, Priddis et al., 2010). When involvement and communication was present, fathers formed a team-like relationship with healthcare professionals. This created an environment in which fathers were not only an integral member of the family, but in which they felt able to express their concerns, fears and feelings, diminishing this sense of loneliness.

2.2.5.3 Working fathers; role conflict

This theme related to the additional challenges and rewards experienced by fathers who worked and how they conveyed their difficulties in balancing their role at home with responsibilities at work. Many fathers felt that it was their responsibility to earn money for their families, with some describing the financial "worry" or "burden" as falling to them to address. This was in part discussed in relation to gender role ideologies and in part, in relation to what worked best for individual families. Whether work was a choice or a perceived obligation, there was a sense of responsibility for fathers to go to work, focus on their jobs, earn money, and then return home to fulfil their role as a father.

"There are often other children in the family to care for, houses to be maintained, employment to attend to" (Jones and Neil-Urban, 2003) (cancer, US).

"Our routine is if he's in a local hospital, which would be great, my wife stays with him at night. When I wasn't working, I'd go in at 7 o'clock or 6 o'clock. She would go to work. Another family member might come in and relieve me. Then she'd come back after work, and then she'd go to dinner and stay overnight again" (Appelbaum and Smolowitz, 2012) (cerebral palsy, US).

"I go to work, I stay at the hospital with my child at night, and then I go back to work" (Chesler and Parry, 2001) (cancer, US).

For some this was manageable, and work provided a respite function and an opportunity to discuss other things (Chamberlain, 2007, Hill et al., 2009). For others, finding this balance was overwhelming and fathers often found it difficult to focus at work and vice-versa, sometimes being tired and unavailable for their families at home. As discussed earlier, fathers often felt like observers in their child's illness and treatment, and workplace factors contributed to this. For example, some fathers were not able to attend their child's appointments due to workplace commitments and felt disconnected from their families. Furthermore, this made building relationships with the staff providing their child's care very difficult.

"Yet another father sadly revealed that he had not been able to attend any appointments with his child for more than a year" (Rivard and Mastel-Smith, 2014) (genetic conditions, US).

Many fathers took on active caregiving roles when they were at home, not only for their unwell child but often for their other children. Furthermore, they also acted as respite for their partner. This could become overwhelming, and fathers portrayed an underlying fear of job loss. Workplace productivity was a concern as many were unable to focus on work following particularly difficult or tiring times at home. Similarly, overload could cause fathers to feel tired and unavailable for their families at home, contributing to feelings of isolation and guilt.

"Juggling home routines, hospital visits, and work responsibilities, some fathers experienced a decrease in workplace productivity and in several cases, employer sanction or dismissal resulted" (Nicholas et al., 2009) (cancer, Canada).

"I'm mostly just there to play with [the ill child]…Sometimes I feel guilty if I have a bad day at work or something like that, and she wants to play, and I'm tired or stuff like that, I sort of feel guilty in the back of my mind" (Nicholas et al., 2016) (life-limiting conditions, US).

This sense of disconnect and isolation was highlighted when fathers were unable to discuss their child and home life at work.

"I'm in sales. Do you think my customers want to hear "Well gee, my daughter's got a brain tumor"? So I keep that inside–so out of my 200 customers, maybe 12 know about her" (Jones and Neil-Urban, 2003) (cancer, US).

Having supportive employers and colleagues who granted fathers flexible working schedules and time off when needed, appeared to make things easier for fathers, not only in terms of productivity at work, but in being able to balance life in a way that worked for them, hence adding an element of control to an uncertain situation. Furthermore, it made work a more pleasant place where fathers could maintain some 'normality'. The value of this in making them feel understood, valued and in control was evident in fathers' accounts of both positive and negative experiences of workplace support:

"However, in those cases where employers did not permit men to take medical (or other) leave to care for their children, fathers were faced with a job loss, change, or move in the midst of the children's illnesses" (Chesler and Parry, 2001) (cancer, US).

“Flexible work arrangements and general workplace support were key factors that helped these fathers adjust to increased caretaking demands” (Brody and Simmons, 2007) (cancer, US).

2.2.5.4 Defining a new normal

Diagnosis marked a very sudden and shocking transition for fathers, with new roles and responsibilities within their families becoming their ‘new normal’. For many of the fathers in these studies, this initial phase was devastating; the futures that they had imagined for themselves and their families would not be fulfilled, and their child would die prematurely. This affected all aspects of their lives. For those children diagnosed with conditions for which there was a potential cure, this shock was no less pronounced, and their families’ futures were filled with uncertainty, compounded by the fear of their child dying prematurely. These changes to expected lives, and in some cases their “expected children”, caused fathers sadness and a new and uncertain future to come to terms with.

“Having a child with CHD meant letting go of expectations and accepting that they would be somewhat different to healthy children. Kevin talked about his assumptions of the ‘perfect pregnancy’ and was aware of lost opportunities like physically bonding with his baby: “What parents will do normally is pick them up and hug them, show them, look here’s the bonding and here we are. And to not be able to do that…”” (Gower et al., 2016) (congenital heart defect, UK).

“Their expectations were of a normal child, a child who could play ball, run, fool around. I’ve never imagined I’d have a son with a degenerative disease” (Lucca and Petean, 2016) (Duchenne Muscular Dystrophy, Brazil).

The process of mourning the loss of their ‘expected child’ and coming to terms with the loss of hopes and dreams they had for their child was an important process. Although it was a time filled with sadness, they were grateful for their child, and redefining expectations helped with their adjustment and acceptance.

"She'll go to school like normal children do but she'll be a bit more breathless and maybe not able to run as fast" (Gower et al., 2016) (congenital heart defect, UK).

"It's just been a heck of a ride. But I'm very grateful for the fact that she is still alive today, and she's a beautiful young lady" (Appelbaum and Smolowitz, 2012) (cerebral palsy, US).

Fathers' lives were transformed in many aspects. They experienced common worries faced by many parents but heightened in the context of their child's condition. For example, they worried about their child fitting in, being accepted, and making friends. Unlike other parents, their lives became consumed with fighting or "battling" their child's illness (Bruce et al., 2016, Jones and Neil-Urban, 2003, Lucca and Petean, 2016, Davies et al., 2004, Hill et al., 2009). They described it as a constant presence from which there was no escape for their family.

Fathers discussed the transformative nature of illness on their relationships with their immediate families. The demands that their child's illness had put onto their relationships with their partners were multifaceted. As discussed, many felt responsible for their partners wellbeing and prioritised their needs, resulting in loneliness and isolation. Challenges arose in response to the huge demands being placed on these families. Relationships became practical, with many couples only seeing one another whilst swapping in and out of their child's care at hospital (Bailey-Pearce et al., 2017, Chamberlain, 2007, Nicholas et al., 2009). However, in contrast, some fathers described their relationships as growing stronger, with many fathers expressing gratitude for and pride in their partner (Appelbaum and Smolowitz, 2012, Clarke, 2005, Chamberlain, 2007, Hill et al., 2009).

"The participants talked about their relationships with their child's mother or partner and in all cases this relationship seemed to have been intensified as a result of their child's illness: 'God knows I wish it hadn't happened but it has made our marriage stronger, much stronger'" (Ware & Raval, 2007) (life-limiting condition, UK).

Relationships with extended family members and friends were also affected, often with huge shifts in fathers' social circles. Much of this was related to fathers seeking those with a shared understanding of his situation i.e. support groups. Fathers described friends as being supportive but not fully understanding their situation with an underlying expectation that their child would get better and hence a decrease in support over time (Nicholas et al., 2009, Chamberlain, 2007, Chesler and Parry, 2001, Hill et al., 2009).

Although fathers had entered this new phase of life, an unexpected and uncertain one, many of them described positive transformations with regards to their outlook on life. Many worried less, focused on the present, lived day-to-day, appreciated what they had and discovered new friendships and organisations. Some discovered the meaning of community after having received financial, emotional and practical support from their local communities.

"For many fathers, this was a source of gratification and vicariously yielded a sense of appreciation for this outcome, despite their difficult and often devastating experiences" (Nicholas et al., 2009) (cancer, Canada).

"Two other fathers shared this view and reflected on how their priorities had changed: None of us might wake up in the morning… that's why we said we don't care what we do or don't do, [we take a] holiday once a year with the two kids. No I never work longer hours, life is too short" (Hayes & Savage, 2008) (cystic fibrosis, Ireland).

2.3 Discussion

Fathers experienced a great deal of uncertainty in the period leading up to their child's diagnosis. This was amplified by a lack of information from healthcare professionals, and fathers described feeling scared and helpless. Elements of adjustment and acceptance followed their child's diagnosis, though the extent to which varied between fathers. Fathers described themselves as supportive figures who often put the needs of their partner and child/ren before their own. This was often mentioned in relation to gender norms and included the suppression of their own emotions out of fear that this would cause distress for others. The way in which professionals engaged with fathers further validated their perceptions of being strong and protective figures. Working fathers struggled to balance their roles at home and at work, and a lack of workplace support created extra stress for these fathers. They discussed the life-changing nature of their child's diagnosis, an event that affected all aspects of their lives, from everyday activities, to their relationships, spirituality, values and ambitions.

2.3.1 Caregiver uncertainty

Caregiver uncertainty is reflected in the wider palliative care literature, both in adults and paediatrics (Choi and Seo, 2019, Arias-Rojas et al., 2019, Snaman et al., 2019) and more specifically in parent caregivers of children with a life-limiting or life-threatening condition (Collins et al., 2016, Bally et al., 2018). Illness uncertainty has been described as "the appraisal of illness and its treatment as ambiguous or unpredictable, or feelings of having insufficient information to cognitively organize the illness event" and has been associated with psychological distress in parents of children with chronic conditions (Perez et al., 2020). Fathers described their uncertainty, particularly at the time of diagnosis, and partly attributed this to a lack of information from healthcare professionals. An individual's ability to manage uncertainty has been shown to impact how they cope with a situation (Einstein, 2014).

Studies exploring the way in which parents cope with the uncertainty, amongst other factors, associated with their child's condition, suggest that they employ a range of strategies, including the suppression of emotions, seeking support, and seeking information (Verberne et al., 2019). In this study, fathers aimed to manage their uncertainty by asserting some control over manageable tasks, such as information seeking and documenting events, which is reflected in existing parental literature (McGeehin Heilferty, 2018). This is also consistent with literature on coping (Lazarus and Folkman, 1984). This pursuit of information is described as problem-focused approach, and is reflected in paternal research in NICU settings (Lian et al., 2020).

However, fathers' informational needs were not always uniform. Some fathers wanted more information than others, with the latter preferring to know as much as they had to in order to get by. Parental literature also suggests that some parents prefer to focus on the "here and now" without detailed prognostic information, and often used this as a means of preserving hope (Verberne et al., 2019, McGeehin Heilferty, 2018). This stresses the need for information to be tailored to the needs of individuals and for accessible information, and preferences that may change over time (Koch and Jones, 2018).

Uncertainty in paediatric palliative care is not unique to fathers, and coupled with a loss of control, appears to dominate many families' experiences (Mitchell et al., 2019b). The way in which fathers managed this uncertainty did not differ significantly to what is represented in existing parental literature though is clearly influenced by many individual factors. However, it emphasised the distress and helplessness that uncertainty can cause and the means by which fathers may feel empowered. Existing studies recommend targeting this uncertainty as a means of reducing psychological distress (Perez et al., 2020).

2.3.2 Engaging and supporting fathers

Fathers' experiences of grief, shock, devastation and sadness are synonymous with those described in wider maternal and parental palliative care literature (Verberne et al., 2019, Koch and Jones, 2018, Courtney et al., 2018). However, this study emphasised fathers' gendered ideas of emotional expression, their minimal outward display of these emotions and the satisfaction of the 'strong and stoic' sociocultural idea of masculinity. Some explicitly linked their emotional expression tendencies to masculinity and 'male roles', whilst for others, it was linked to a more subliminal desire to protect their families from further emotional distress. Regardless of how gender identity appeared to shape these accounts, this internalization of distress and lack of expression, alongside this perceived obligation to care for others, are indeed rooted in traditional ideas of masculinity (Knights, 2019). Not so clear are the differences between fathers for whom performing this supportive role was helpful and for whom it was fulfilled out of perceived obligation.

Putting others' needs first often resulted in fathers being secondary figures in the context of their child's care. For some, this was presented as an active and conscious choice. However, there was also a level of dissatisfaction amongst fathers who were left feeling like they were observers in their child's care. This peripheral role seemed to be reinforced by fathers' interactions with professionals, who often interacted with mothers more often than them. Furthermore, fathers were rarely addressed regarding their own support needs, further validating their perceptions that their emotions were not relevant or necessary to share. Many fathers discussed the eventual need to discuss their emotions, with some wishing that they had sought more support in the early days of their child's illness.

This issue is reflected across paediatric healthcare settings and literature on healthcare structures though father involvement is growing, particularly in NICU settings. The literature illuminates some of the challenges and benefits of, as well as strategies for, improving father involvement and engagement (Kim et al., 2020, Hearn et al., 2020). Fathers have been shown to seek help, participate in decision making and contribute to their child's care when they feel accepted and comfortable enough to be assertive. This ability may be hindered if fathers are on the periphery and are expected to act only as a support for others; it is particularly hard to engage fathers as caregivers when they perceive their role to be a supportive one (Pfitzner et al., 2018).

Communication challenges are not unique to fathers in this context, with similar findings reflected in existing parental literature (Bally et al., 2020). However, these findings highlight fathers' communication needs as well as their experiences of poor communication and the effects of such. They demonstrate that fathers do want to be recognised and actively involved in their child's care. The difficulty lies in creating an environment in which nuanced family dynamics can be recognised and accommodated. Fathers are not a homogenous group of individuals with the same experiences, though it is clear that there needs to be a deeper appreciation for the benefits of effective professional engagement and accessible support and information for this group.

2.3.3 Workplace

The role conflict experienced by working fathers highlighted several unique challenges faced by working fathers. There were few fathers in this study who assumed the role of primary caregiver for their child, with the majority being in employment, following the traditional role division that still exists more generally (Schneider et al., 2011). However, many fathers expressed a desire to be more heavily involved in their child's care, which was sometimes made difficult by their workplace responsibilities. Studies show that employees with caregiving responsibilities are at a higher risk of negative workplace outcomes, such as dismissal (Brannan et al., 2018, Halinski et al., 2020) which was demonstrated by fathers' anxieties related to their performance at work. These fathers faced problems in attending their child's appointments, leaving them feeling disconnected from their families. Research also shows that limited uptake of additional paternity leave (APL) is in part due to concerns over a lack of employer support (Kaufman, 2018) which further highlights struggles faced more generally by fathers at work. Existing research focuses heavily on female caregiver discrimination in the workplace (Bose et al., 2020), meaning that not much is known about problems faced by men when they have caregiving responsibilities, particularly fathers of unwell children.

Conversely, fathers discussed flexible working and positive employment experiences. For these fathers, their employers made it possible for them to attend appointments, work from home and take leave when needed. Flexible working policies have increased across the workforce in recent years (Chung and van der Lippe, 2020) and allow for individuals to balance their work-home roles more easily. However, access to flexible working arrangements are not uniform and are affected by occupational group which may explain the polarity of the working experiences in this study (Chung and van der Lippe, 2020). Other positive experiences related to fathers' work serving as a respite function, allowing them to maintain a sense of self. These positive *and* negative experiences emphasise the importance of workplace support for parents and the recognition that more fathers want to be involved in the care of their children.

2.3.4 Altered life

Fathers described their lives as being completely altered by their child's diagnosis. This is consistent with existing parental literature that describes parents as "travelling a different pathway" to the expected (Collins et al., 2016). For many in this study, life became about "battling" their child's illness, leaving little time for anything else which is also demonstrated in existing literature on both parents (Postavaru, 2018). Day-to-day living was often consumed by the illness, with fathers describing their hypervigilance and exhaustion. Fathers described shifts in their relationships, both with their partners/wives and their wider groups of family and friends, some becoming difficult due to a lack of understanding. A loss of common interests with friends and withdrawal from existing social circles and a shift towards those with a shared understanding has been described (Rodriguez and King, 2009). In this review, fathers described a sense of loneliness in their experiences, though the desire to talk to others was mixed. Some found solace in support groups of parents of children with similar conditions and found new meanings in community.

2.3.5 Strengths and limitations of included studies

There were numerous strengths to this review. I included 32 papers from 30 studies, containing the experiences of 576 fathers across multiple countries and settings. I drew my findings from many perspectives, allowing the exploration of the unique experiences of fathers. In choosing thematic synthesis as an approach to analysis I was able to include studies from a range of methodologies, settings and diagnoses. As far as I am aware, this is the first review on this area that includes father-only studies. A meta-ethnography of fathers' experiences of having a child with a life-limiting condition was published shortly before this study was completed (Postavaru et al., 2021). However, that review did include studies that had also sought the perspectives of mothers which was a key exclusion criterion in this review.

Only studies published in English were included as it was beyond the scope of the review to include studies published in other languages. Fifteen of the included studies were focused on the experiences of fathers of children with cancer. Furthermore, studies that included a broad range of life-limiting conditions also included fathers of children with cancer, meaning that there was further bias towards oncology settings. In an attempt to assess the impact of this on synthesis, I coded studies looking at the experiences of fathers of children with cancer last. There were not any substantial additions to the coding structure at this point, suggesting that the experiences of fathers of children with cancer share some key similarities with those of fathers of children with other life-threatening or life-limiting conditions.

The majority of the studies were published in the US where healthcare structures differ significantly. This is particularly important when we consider fathers’ experiences in struggling to pay for medical bills and subsequent financial worries. Studies were published between 1997 and 2019 and data was collected between 1978 and 2019 providing experiences that span many years, across changing healthcare systems, medical developments, and societal views. There was a lack of cultural diversity amongst the included participants as well as a lack of consideration of the inequity that exists in relation to access to palliative care, including the multiple factors contribute to this inequity.

It was difficult to assess differences between other groups of diagnoses as some groups were only included in one or two papers. It is also important to note that there were differences in the relationships with staff in hospice and in hospital settings. Relationships with healthcare staff at diagnosis were often difficult due to a lack of communication and trust. Those under the care of a hospice discussed relationships in more positive light suggesting that hospice staff were more attuned to family-centered care. Palliative care clinicians have been shown to assess understanding of disease more so than oncologists (Thomas et al., 2018).

2.3.6 Strengths and limitations of review methods

I sought identify existing literature surrounding the experiences of fathers of children with a life-limiting condition. The search strategy was extensive and allowed for the identification of 32 papers from 30 studies. Given the increase in the prevalence of children and young people with a life-limiting condition, it is important to understand the experiences of their families who often provide all of their care. This includes an increasing number of fathers, who have often been excluded from research. This review is unique with respect to the included studies, which focus solely on the experiences of fathers, rather than mothers and fathers. Existing reviews that explore fathers' experiences have generally included mothers (Sjuls et al., 2023). The method of synthesis, thematic synthesis, allowed for a rigorous analysis and the inclusion of a studies from a range of methodologies, settings and diagnoses.

2.4 Conclusions

Fathers have been under-represented in paediatric palliative care research. This review has highlighted that fathers’ experiences share some key similarities with those reflected in the wider maternal and parental literature. However, their unique experiences as fathers should be considered in order to provide a family-centered model of care. Fathers describe their experiences as the ‘side-lined’ parent in healthcare settings and their conflict between roles at home and at work. Workplaces should strive for flexible policy that allows for both mothers and fathers to be involved in their children’s care. Further research is required to understand the nuances of fathers’ work and home role balance.

Fathers experience many negative emotions in response to their child’s diagnosis and ongoing treatment. Their responses are not widely understood, but their exposure to numerous risk factors for poor psychological outcome warrants further study. They describe exhaustion and hypervigilance, role overload and a lack of support, which are all risk factors for depression and anxiety (Li et al., 2020). Whilst this study was able to shed light on the broad range of experiences of these fathers, studies only briefly, if at all, touched upon the impact of these experiences on fathers' health and wellbeing. Furthermore, due to a focus on distress, the positive aspects of caregiving and impacts on physical health were not covered. Given our improved understanding of the impact of caregiving on the physical and mental health of those providing care, further research is warranted to understand fathers in this context.

3. The health of mothers of children with a life-limiting condition: a qualitative interview study

My PhD sits within a wider study: Crisis prevention rather than crisis management; the health of mothers of children with a life-limiting condition. Alongside my PhD, I was employed as a research trainee on this wider study. In my role, I was responsible for the general running of the qualitative aspect of this study, including liaising with research sites and clinicians, developing and distributing study documentation, liaising with our PPI group, interviewing mothers, and analysing the findings. The core design of this study had already been established and approved by an ethics committee prior to the commencement of my employment. The overall study was made up of multiple workstreams including both quantitative and qualitative components and, as mentioned, I was involved in the qualitative component; semi-structured interviews with mothers of children with a life-limiting condition to explore their health and experiences.

As the mothers' study progressed, it became clear that it could make a valuable contribution to this thesis, alongside the work I was doing with fathers. The focus on the experiences and health of mothers, including how they think their health should be supported, generated important questions surrounding the health of fathers. Although we know that mothers are more likely to be their child's primary caregiver, the findings of the systematic review in chapter 2 highlighted the dearth of evidence in relation to the impact of caregiving on the health of fathers. Contemporary studies have rarely covered parents' perspectives of their health, nor any differences in the experiences of mothers and fathers in this context. The inclusion of the maternal health data provides the opportunity for comparison between two datasets, exploring issues that have so far not been covered in detail in this population.

The data collection and analysis process in this study provided me with the opportunity to develop my own skills as a researcher prior to undertaking the work with fathers. Furthermore, it provided the opportunity to make comparisons surrounding data collection methods, what worked well for mothers vs. fathers and to contribute to discourse surrounding the recruitment of fathers more generally. This will all be explored in detail in the discussion in **chapter 6** alongside how the findings of this study relate to those of fathers.

Although this study is presented before my methodological rationale for the primary study on fathers, there are important points in chapter 4 that are relevant to how I carried out the maternal health study data collection and analysis, including epistemological considerations and reflexivity. In such cases I will reference subsequent sections that can be found in chapter 4.

This qualitative study has been published (see Appendix 4):

Fisher V, Atkin K, Fraser LK. 2022. The health of mothers of children with a life-limiting condition: A qualitative interview study. *Palliative Medicine,* 36(9), 1418-1425.

3.1 Background and rationale for study

As highlighted in chapter 1, as the number of children with life-limiting conditions increases, so do the number of parents providing care at home (Fraser et al., 2020b). Evidence tells us that mothers of children with a life-limiting condition are more likely to experience poor physical and psychological health outcomes than other mothers, including back problems, heart problems, arthritis, anxiety and depression (Fraser et al., 2021). Although there is a growing body of research surrounding mothers' experiences and support needs, information about the impact of caregiving on their health specifically, as well as how, and by whom, mothers feel that their health concerns should be best managed, is rarely discussed. Existing studies usually explore parental health in the context of special needs or specific conditions, meaning that we know little about health issues may manifest in mothers of children with a life-limiting condition (Fraser et al., 2021), This re-emphasises the need for further research on appropriate physical and psychological support for these mothers, also highlighted in National Institute for Health and Care Excellence (NICE) guidelines (NICE, 2016). This is important not only for mothers themselves, but in ensuring that they feel able to care for their child. This study addresses this gap by exploring mothers’ accounts of their mental and physical health; their experiences of accessing healthcare; and views on which services should support their own healthcare needs.

3.2 Methods

3.2.1 Participants

The inclusion criteria for the study were as follows:

- Mothers of children with a life-limiting condition.
- Mothers aged 18 years or over.
- Mothers of children (aged 25 years or under) with a life-limiting condition or whose child had died from a life limiting condition (at aged 25 years or under).

Mothers were excluded if:

- They were aged less than 18 years.
- Their child had died in the last 3 months or longer than 3 years ago.
- They lacked capacity to participate in the study, guided by the 2005 Mental Capacity Act.

3.2.2 Recruitment

Participants were recruited via three UK-based children's hospices and via social media (Twitter, Facebook) between November 2020 and June 2021. It is important to note that this was during the COVID-19 pandemic. Purposive sampling was undertaken by healthcare professionals during consultations, meetings, and visits, as per the study inclusion criteria. Healthcare professionals provided eligible mothers with the study information, following which potential participants gave written consent for their details to be passed onto me. I contacted all participants who had completed the consent-to-contact form through their preferred contact method. I discussed the study with potential participants, checked their eligibility and answered any questions they had. We then booked an interview if appropriate. I received 39 consent-to-contact forms. Potential participants who saw the study advertised on social media were directed to contact me directly, following which the same steps as above were followed.

3.2.3 Data collection

Semi-structured interviews were conducted between November 2020 and July 2021. The topic guide (appendix 5) was developed from the literature and then refined with a patient and public involvement (PPI) group and through piloting, prior to my involvement in the study. Given the qualitative and exploratory nature of the study, the topic guide included various topics surrounding the broad exploration of mothers' own health concerns, ease of access to healthcare for themselves, ways in which they look after their own health and suggestions on which services should look after their health. 30 interviews were carried out, and I conducted 15 of these myself. Following verbal or written consent, interviews were carried out on the telephone or via video call, depending on participant preference. We were unable to conduct face-to-face interviews due to the COVID-19 pandemic. Probes and additional questions were used for clarification and deepening of understanding. Interviews were audio-recorded, transcribed verbatim and anonymised prior to analysis. It was not possible to conduct face-to-face interviews due to restrictions put in place during the COVID- 19 pandemic. The potential impact of this will be discussed later in this chapter and in the discussion in chapter 6.

3.2.4 Ethical considerations

Ethical approval was obtained for the study from the North East – Newcastle & North Tyneside 1 Research Ethics Committee on 2nd July 2020 (REC reference 20/NE/0164- see Appendix 6). As mentioned above, potential participants were informed about the study via social media or were approached by staff at participant identification centres (PIC's). Potential participants were given the full information sheet for the study before taking part in the survey. Participants provided written consent for the survey. If they decided to take part in interview as well, they were given another information sheet detailing the interview process further. All participants gave written or verbal consent prior to the interview. Participants were given the opportunity to ask any questions about the interview and were given time to consider participation. I made it clear to participants that they could stop the interview or withdraw at any time, without giving reason. I also explained that the interview was confidential, unless they told me something that caused concern in relation to their own, or someone else's, safety. I explained that I would discuss this with them if that happened. If participants became distressed during the interview I asked if the participant would like to stop the interview, take a break, or rearrange it. Names and other identifying information, such as specific hospitals/wards, were redacted from the interview transcripts. Ethical concerns remained surrounding the identifiability of participants given the rarity of some of the conditions included in the study. Therefore, specific conditions were also redacted, and participant characteristics were presented as group variable summaries. A broader exploration of ethical considerations is included in chapter 4, relevant to both mothers and fathers.

3.2.5 Analysis

I organised, managed and coded all interview data in NVivo and to analyse the data I used reflexive thematic analysis (Braun and Clarke, 2006, Braun and Clarke, 2020, Braun and Clarke, 2019, Braun and Clarke, 2024). The stages of, and rationale for, using thematic analysis are covered in chapter 4 and are also applicable here. To summarise, reflexive thematic analysis involves; (1) familiarisation through reading and re-reading; (2) generation of initial codes; (3) theme development; (4) reviewing of themes; (5) defining and naming themes; (6) writing up and summarising themes. Data analysis was approached inductively and iteratively. I led this analysis with frequent discussions with the wider research team and my supervisors.

3.3 Findings

Thirty mothers completed interviews; 24 were recruited via children's hospices and six via social media. 39 consent-to-contact forms were received from children's hospices meaning that 15 mothers decided not to participate in the interview after providing their details for me to contact them. For some, this was due to a lack of time to participate in an interview and for others, contact was lost. Mothers were aged between 32 and 60 and resided across various regions of the UK. The mothers of 34 children were interviewed as some participants had more than one child who had been diagnosed with a life-limiting condition. The characteristics of the children are shown below in table 5. Interviews lasted between 72 and 102 minutes (average 63 minutes).

Table 5; child characteristics

Characteristic		
Sex of child	**n**	**%**
Male	16	47.1
Female	18	52.9
Age of child		
0-5	7	20.6
6-10	14	41.2
11-15	4	11.8
16-20	5	14.7
21-25	4	11.8
Mean age 10.8 years		
Child's diagnosis		
Neurological condition	9	26.5
Genetic condition	19	55.9
Congenital condition	1	2.9
Metabolic condition	4	11.8
Cardiac condition	1	2.9
Age at diagnosis		
Before birth	1	2.9
At birth	4	11.8
Infancy (0-1 years)	11	32.4
Childhood (1-9 years)	13	38.2
Unknown	5	14.7

Four analytical themes were identified during analysis which are detailed below. Together they describe the experiences of mothers, related to their own health and experiences of caring for a child with a life-limiting condition. The themes detail health problems experienced by mothers, things that help them to look after their health, barriers to support, and the impact of ongoing battles with services.

3.3.1 Theme 1: The unique experience of parenting a child with a life-limiting condition: A need for understanding

This theme described the unique experience of caring for a child with a life-limiting condition. It highlights the complexities associated with such, not only in terms of the complex medical routines, but in relation to the impact that such has on mothers themselves. Mothers felt that their experiences, and often the extent of caregiving, could go unnoticed, highlighted by what they considered to be a lack of appropriate understanding and support from healthcare professionals. They described a healthcare system in which they found it challenging to seek timely care for themselves alongside prioritising the healthcare needs of their child. This was prominent in their experiences of seeking support through their GP.

Physical health problems were prominent in mothers' accounts of their own health, mainly in relation to the effects of lifting/carrying their child. As well as the physical effects, mothers described their experiences of anxiety, depression, and post-traumatic stress.

"It had reached points where I couldn't even stand in the last 8 months but it has been virtually impossible to get a GP appointment. Because I have to phone to renew [child's] medication, if I can get through to reception, that's my priority. It has taken 28 phone calls on occasion to get through to anybody" [Mother 2].

For some these symptoms/conditions were fairly constant, sometimes expected if mothers had a history of mental health issues, whereas for others the onset of such could be sudden and unexpected, having never experienced mental health problems before.

"I went from coping with everything to just not. . .I had zero energy. Just the thought of telling somebody what had happened to [child], I just couldn't bear it. I realised that it was depression. That was very unfamiliar to me" [Mother 25].

For some this meant it could be difficult to seek support/care. A key issue for many was poor access to timely healthcare followed by inappropriate treatment or referrals being offered. This was particularly the case for mental health concerns; prescriptive therapies with a limited number of sessions, such as cognitive behavioural therapy (CBT), were perceived to be unhelpful in their scope in terms of both content and number of sessions available.

"I went to the first CBT appointment. I spent 15 minutes going through everything, past, present and future, and they kind of sat there and their jaw got progressively lower and lower. At which point they literally went 'I'm going to stop you there because this is well beyond what we can deal with. We're only given funding for 12 weeks and this is a lot more than 12 weeks of stuff" [Mother 8].

"Well, because I think you don't always, you don't always, it can feel a bit forced because especially if it's just … it was very different when I actively depressed but otherwise if you're just having a bad time it can feel a bit forced and like you have to go and have something to say (laughter) which you don't always and also like the idea of having something in eight week blocks and then you're better and if it's a kind of life event I think it's more, it's not going to get better" [mother 1].

There was a real sense that psychological therapies, and the providers of such, were unable to recognise, or address, the true nature of providing complex and extensive, often life-saving, care for a child.

"You say, "disabled child" and people do not think what sort of child I have, so yeah... you can't really have counselling and not have an understanding of that part of my life, like for my anxiety for example it's very much for me like a health anxiety for me and about very specific things; I think people that can understand that can understand the level of anxiety and worry and everything that goes with having a child like that get all that" [mother 27].

Furthermore, mothers struggled with such professionals understanding of the impact of such care.

"I think I struggled with the concept that my thought processes were irrational. I think I kind of looked at our circumstances and thought "to be honest most people would feel a bit crazed" and I really struggled with that" [Mother 3].

There was a real concern from mothers that self-perceived normal reactions to extremely challenging situations were automatically labelled as a mental health issue by professionals. They found this unhelpful and believed such responses further failed to recognise the realities of caring for a child with complex needs.

"I didn't feel like I did have anxiety or depression. I suppose it is anxiety, but it's a different anxiety when your child stops breathing, it's reasonable anxiety" [Mother 19].

"I don't know, it was hard, there was a lot of emotion but I don't know if I would say that was a mental health thing, it was a: I have a really sick baby thing" [mother 28].

Mothers also found that such support was often offered at inappropriate times of day i.e. when they were unable to attend due to their caregiving responsibilities, corroborative of the idea support was inappropriate for them as caregivers.

"I'm not even sure what her job title is, I think she's, like, a counsellor. So, she was involved. I think I could just ring up and book a session but at the minute I just find that I'm, sort of, too busy, most of the time" [mother 4].

One mother had found CBT to be very helpful in addressing her anxiety, particularly in understanding the underlying physical mechanisms of anxiety and individual coping responses to such. Importantly, in this case CBT was delivered in combination with additional support from a children's hospice, focused upon the specific impacts of caregiving.

"But the other stuff has been really helpful, particularly the CBT with my anxiety was very helpful, understanding what was going on, the physical stuff that was going on in my body, I'm really like an information person so if I can rationalise things, I understand what is that feeling that really helps. I felt like she armed me with information but also challenged some of the coping mechanisms I'd given myself that weren't very helpful but you don't even realise you're doing really at the time. And then with the hospice support that's been more focused

on [child] and sort of the effect I guess he has on us, so a different focus but also very helpful" [mother 28].

3.3.2 Theme 2: Mothers' reluctance to prioritise their own health and needs

The first theme explored difficulties associated with accessing timely and appropriate support, with mothers describing a lack of recognition of their individual needs and a subsequent disengagement in addressing these needs. However, analytically distinct from this first theme was a deeper sense of unwillingness to prioritise their own needs, both in terms of healthcare and general support. This theme is broken down into three sub-themes contributing to this reluctance; a lack of trust in others' ability to care for child, worries about judgement from others and a need to be strong for others.

3.3.2.1 A lack of trust in others' ability to care for child

Mothers described having an instinct for their child's needs and felt they were the only ones able to provide adequate care for them. Mothers described their fear of handing over their child's complex care routines to others. This was due to their inherent lack of trust on others' ability to carry out this care, and sometimes due to previous experiences of problems related to handing over care. This meant that it was difficult for mothers to ask for or accept help, whether they felt they needed it or not, and therefore breaks from caregiving were rare.

"I don't trust family to look after [child]. Not that I don't trust them, I don't trust their medical ability to cope in a crisis. It's difficult and incredibly stressful and takes a lot of inner strength to cope with it". [Mother 25]

Leaving their child with others could cause great anxiety for mothers. Even respite support provided by children's hospices was often difficult for mothers. This was not so much related to a lack of trust as it was to a fear that their child's condition could deteriorate, and they would not be there. Another issue with handing over care was that mothers could find it difficult to explain their child's complex care routines/medication routines to others. Preparing their child for time away from them could be emotionally and practically demanding.

This lack of trust in others, and other worries associated with handing over care, led to concerns that if mothers themselves did become unwell in the future, their child would not be looked after properly. This generated future anxieties and vulnerabilities and may also explain why some ignored initial signs of their own illnesses.

"I can't even go there yet. Like I didn't think she had years to come. But, you know, yes, in the bigger picture, I need to be in the full of my health, and I feel quite vulnerable, and I'm really conscious that I don't have a sense that I can look outwards [tearful], you know. And this is partly me trying to break through that and daring to say it to a doctor, going – I really am terrified of doctors now". [Mother 17]

There was, however, a level of recognition that mothers needed to be well to provide the care for their child, particularly for those who had experienced issues with their own health in the past. For some this manifested as making sure that some self-care was observed, although as we have seen the demands of care can make this difficult.

"To be able to look after [child], I need to look after myself. It's a full-time job and when things go wrong, it happens quickly. So I just need any support I can get and to look after myself so that I can cope when that happen"'. [Mother 25]

3.3.2.2 Worried about judgement from others

Another barrier to mothers seeking support originated from the concern that if they asked for help, they would be judged for '*not coping*' or else be perceived as a burden to others. This was touched upon in the first theme surrounding healthcare professionals misunderstanding the experiences of mothers. However, this sub-theme is more specific to feeling negative judgement from others, including healthcare professionals.

"I spend a lot of time worrying if I have said the right thing [to healthcare professionals], whether it will affect how they judge us as a family and support us as a family" [mother 4].

There was the sense that mothers could handle having 'bad days', and that they were a normal part of life. However, mothers did not want others to witness such days.

"I don't want people to worry about me and if I have a really bad day, I don't think I'm able to call anybody, I just want to crawl away and wait until it's over and don't want to worry people". [Mother 12]

Some mothers felt that they could manage any mental health difficulties, with the prospect of others' judgement the most difficult aspect of such.

"Once he was born...it was really hard but I think one of the things I struggled with early on is that I didn't have, I'm still like this a bit that I don't … I guess some of my anxiety and things I get anxious about and stuff started to play into it in terms of how we managed hospital stays with him and it was fine for us, we managed it fine as a family but then I started to second-guess what people thought" [mother 28].

This fear of judgement was closely linked to mothers' perceptions of what it meant to be a good parent. However, mothers did highlight that the way in which they portrayed themselves as resilient had consequences in subsequent responsibilities placed upon them as caregivers e.g. *"the more I do, the more they think I can do. I'm doing it because I don't have a choice but because it looks like I'm coping, I get more and more offloaded onto me"* [mother 25]. This reinforcement of the 'capable mother' construct meant that mothers found it even harder to ask for support, out of fear that they would be letting people down. Furthermore, mothers felt that services could take advantage of this so called resilience.

"And I think actually it was a mistake to come across as that resilient and capable because people think now they can push it to insane levels. If I was a qualified nurse I would not be allowed to work on the level of sleep and do the interventions I'm doing" [mother 2].

This leads us onto the next subtheme *a need to be strong for others.*

3.3.2.3 A need to be strong for others

Mothers believed that they needed to be strong for other family members. When asked to talk about their own health, mothers frequently went on to talk about what things were like for their child and family. This further highlighted a reluctance to talk about themselves and how closely related their own wellbeing was to that of their child and family. Mothers felt protective of their partners' wellbeing and felt that highlighting their own struggles would make things more difficult for their partners. This was less related to wanting to be *perceived* as strong mothers, than merely wanting to protect their partner from further distress.

"He [partner] struggles as well. If I'm struggling he knows, he just knows I am but I don't highlight it because I don't want to set him off. He had a number of issues over the last 10 years, always around [child's] birthday time". [Mother 1]

3.3.3 Theme 3: Stress resulting from battles with services, rather than as a direct result of caregiving

This theme describes mothers' interactions with services, which they often talked about as 'battles' or 'fights', ongoing for extended periods of time. The nature of the impact of such battles varied, depending on the type of support that mothers felt they were lacking. It could include anything from access to specific equipment to more general support. A key issue for mothers was having to lift their child whilst awaiting appropriate equipment caused back problems and pain which could become frustrating.

"Not having the correct equipment is a big one. Because of COVID things have slowed down. I've waited 6 months for a sling. The sling is the most important thing in our house at the moment. We need it. She's a big girl and as a result of us not having a sling, me and dad have now got back problems. I've been waiting months for a hoist. It was like this even before COVID". [Mother 27]

This was also the case for mothers who described waiting for appropriate housing, where they would not need to carry their child up and down stairs.

"I think we started maybe about three years ago and we've had an architect round, was it just [a month ago]? So, it's slow going. He usually crawls upstairs and he comes down on his bum. But obviously that's not going...There is a lot of lifting". [mother 9].

One issue highlighted in relation to this is that some battles could go on for such an extended time that once families were provided with the equipment they had asked for, their child had outgrown it and an updated version was required, meaning further battles with services. Sometimes families would raise the funds for equipment themselves meaning that financial wellbeing could become an issue. Mothers regarded these 'battles', (particularly for equipment), as relentless and difficult to manage, especially when they had to be negotiated alongside other caring demands. They felt that managing many different challenges had an accumulative impact on their wellbeing.

"I will hold my hands up and say yes, it is hard. But it would be easier if we didn't have to fight for the help that we need and have to chase people who don't do what they say they are going to do. It just adds to the feeling of general tiredness and exhaustion really". [Mother 30]

Mothers highlighted these 'battles' when responding to questions about their physical and mental health. They reiterated that their stresses arose from these interactions with services, rather than as a direct result of caregiving. This further demonstrated how closely linked mothers' wellbeing was, with how well their child's needs were being addressed.

"The frustration isn't medical. The frustration is the network of services. It's the constant battle and how massively time consuming and frustrating it is". [Mother 2]

Mothers found is particularly difficult when their child was denied support from services or access to education. Mothers felt a real sense of injustice related to their child not having access to the support that they deserved. It could become increasingly frustrating for mothers when barriers to support were seemingly created by services themselves.

"[Child] was basically removed from Community Healthcare, removed from education, all our home care was removed when I was pregnant with [sibling], not because anyone thought we

didn't have serious needs but because everybody thought it was somebody else's job" [mother 2].

3.3.4 Theme 4: Establishing connections; feeling recognised

This final theme explores the value of feeling recognised, particularly the importance mothers attached to care professionals understanding their situation. Some of this relates to what was covered in theme 2; the need to be understood by professionals in relation to the impact of caregiving on their **own** health and wellbeing and what appropriate support looks like. Theme 4 combines factors that seem to mitigate negative impacts on the health and wellbeing of mothers as well as factors that seem to actively help mothers to deal with said impacts.

This was often in the context of their child's care, i.e. professionals or processes directly involved in their child's care. Mothers, for example, emphasised the benefit to their wellbeing, of feeling they had established a rapport with professionals involved in the care of their child. Mothers spoke of how such rapport helped mitigate against the stresses associated with providing constant care for their child. It also often helped mothers reflect on and address their own health. Mothers identified hospice support as especially important. They felt able to discuss their own needs with hospice staff, who they believed understood their family and perhaps more importantly, the challenges of caring for a child with complex needs. Mothers expressed frustrations about constantly needing to explain the child's condition (and needs) to other care professionals. This further explains the value of hospice care.

"Having counselling through the hospice. . . firstly they have met [child] which makes a difference. . . it's all one team. . . I know if I had the choice I would always access support through the hospice because it's such a massive part of everything. You can't have counselling and not have an understanding of that part of my life". [Mother 22]

Mothers also specifically valued support offered directly alongside their child's care, knowing that their needs were being taken into consideration.

"Yeah so the NICU. . . basically assume that you need it and want it [counselling], so it's an opt out system rather than an opt in. So the second [child] was born we were assigned a counsellor and within a week she had come to the cot side and introduced herself and said "shall we book a time in another room to sit and talk so we're not talking around an incubator?". It should be provided. I say that very strongly, should be provided". [Mother 23]

This contrasted with mothers' experiences of support from other professionals. When asked 'when was the last time that a healthcare professional asked how you were?', mothers explained that this was not only rare, but carried with it an assumption that they should answer that they were 'okay', as there was not time to discuss any issues during their child's appointments, a failure in recognition that their wellbeing was connected to that of the wellbeing of their child.

"There's not the holistic family approach, it's just focused on [child] and we just have to, you know, do our best, but our wellbeing isn't really considered". [Mother 30]

This reiterates the value of support for parents that was able to be integrated with their child's care. Additional support and activity groups involving other mothers could also be useful, particularly for generating a shared understanding of experience, alongside practical advice related to getting appropriate equipment and support. However, mothers were keen to highlight that some of the groups they attended included parents whose child had different needs to their own. At times, this created challenges in generating a shared understanding and prompted feelings of isolation.

"As soon as you come into the special needs world you are suddenly just surrounded by ADHD and autism to be perfectly honest and that's not us, I have a child who is not even on the autistic spectrum and it's very hard to access services, it's very hard to get anybody to think it matters because a lot of people assume if you've got a serious child, "Well you must get tons of support" And often we get in many ways the least because we can't access universal services like after school clubs or even school a lot of the time". [Mother 17]

This theme highlights the need for mothers to feel recognised in the context of their child's care and as individuals. Services that recognise the challenges associated with caregiving, such as children's hospices, were valuable. It could be difficult for mothers to disentangle and address their own support needs from those of their child, but services practicing with an understanding of this seemed to help to mitigate some of the negative effects of caregiving, giving mothers the space to consider their own needs when they might otherwise have felt a sense of invisibility.

This sense of invisibility seemed to permeate mothers' accounts of caregiving. Together with the other three themes, this final theme has highlighted the extent of the responsibilities that mothers have in providing care for their child. Feeling recognised in this was incredibly important for mothers, whether it was by healthcare professionals or those with shared understanding of their experiences.

I want to finish this theme, and results section, with a quote from one mother who wanted to remain in employment alongside her caregiving responsibilities.

"The thing about work is I feel it gives me some sort of value to the rest of the world and actually what I do is not really very valuable but it's a status thing that's what it is, it's a recognition that people think that I'm talented and I've got something to offer, "Isn't she amazing?" Whereas if I don't have that I just feel invisible basically" (mother 13).

3.4 Discussion

This chapter details a study conducted as part of my role as a researcher alongside my PhD. Data collection and analysis occurred in parallel with the design of the primary study on fathers (chapters 4 and 5) and, as mentioned, it became clear that there were important comparisons that could be made between the experiences of mothers and fathers. This would enable me to identify both similarities and differences in the experiences of mothers and fathers, important in the context of paediatric palliative care and any policy recommendations. Comparisons will be directly addressed in chapter 6, once I have discussed the results of the fathers' study. Below, I will summarise the findings of the maternal health study, discuss how they build upon existing literature, and outline their value in the design, conduct, and analysis of the fathers' study.

3.4.1 Summary of main findings

The aim of this study was to explore mothers’ accounts of their physical and mental health and to identify how, and by whom, their own wellbeing should be addressed. Previous research tells us about the types of physical and mental health conditions present in this population including anxiety, depression, and arthritis (Fraser et al., 2021), but these conditions, the impact of such, nor mothers' own health behaviours have rarely been explored qualitatively. In this study, mothers described a healthcare system that did not accommodate nor recognise their needs as caregivers. They highlighted how care professionals could misinterpret mental health concerns, failing to recognise the unique nature of caregiving and differentiate between normal and pathological distress. This could create further tensions through which mothers felt that they could not express their needs out of concern that it would appear that they were unable to cope.

Mothers also highlighted how their wellbeing was mostly affected through conflicts with services, rather than as a direct result of caregiving. 'Battles' and 'fights' with services for equipment or other provisions could leave mothers feeling drained. Mothers described situations in which it was even difficult to acquire essential day-to-day provisions such as medication for their child, highlighting the constancy of their roles in ensuring their child has what they need. Mothers' positive experiences of support were closely linked with how well they felt their child was being supported and provided for. This created tensions in which mothers needs were strongly linked to those of their child, alongside also wanting to be recognised and supported as caregiver in their own right. This was difficult, but even more so when healthcare professionals did not recognise the complexities of this, meaning that mothers sometimes dismissed the challenges they faced. This created an environment in which mothers were unsure about how to express and navigate their concerns relating to their own wellbeing and worried about being misinterpreted when articulating these concerns. This explained their sense of vulnerability when seeking support.

Related to this was the. value that mothers placed on the practical and emotional support that HCP's *did* provide. Children's hospices were environments in which mothers' own needs could be addressed alongside those of their child, though there was still an underlying reluctance to mothers addressing their needs regardless of what support was available. This was sometimes related to avoidance due to a fear that if they did become unwell, there would be no one else capable of looking after their child. Mothers described a lack of trust in others' abilities to care for their child, particularly when their child required substantial medical intervention and monitoring.

3.4.2 In context with the wider literature and what this study adds

The psychological and physical health problems identified in this study reflect those in the wider literature, including mothers' experiences of anxiety, depression, and stress, alongside physical problems such as back pain and arthritis (Fraser et al., 2021, Collins, 2020). Equally, this study reiterates the issues highlighted around how difficult it can be for parents to obtain appropriate support for their child. "Fighting" or "battling" services is well cited in this context (Mitchell et al., 2021, Price et al., 2022, Rud et al., 2023). However, the impact that this has on the health and wellbeing of mothers, and the extent to which this prevents them from addressing their own health, has not been covered. Navigating a disjointed system and 'battling' for support with little or no means of guidance was challenging and overwhelming when considered alongside the challenges of their everyday caregiving responsibilities. This is important when understanding the context of mothers' psychological health and should be considered alongside mothers' perceived inadequacies of current support when deciding on appropriate support and/or interventions. Focusing on mothers' psychological experiences as requiring professional intervention was problematic for the participants in this study. "Goal-oriented" forms of therapy, such as CBT, seem to sit in conflict with mothers' experiences of caring for a child who is not going to get better. Furthermore, when we consider the brief nature of such therapies, with some mothers reporting as few as 6-8 sessions, it is possible to understand why this is not a sustainable nor useful means of support for these mothers.

Understanding the increased risk of anxiety and depression in these mothers is important, but the context underpinning this data has not been explored until now. Mothers feeling misunderstood, lonely, and vulnerable with limited or erroneous treatment/support options, demonstrates the need for further improved understanding of how these mothers can be supported to stay well. Other studies have begun to delineate the problems associated with a focus on illness and distress, lack of caregiver diversity, and a disregard for positive aspects of caregiving (Greenwood et al., 2018). It could be argued that the lack of exploration of mothers' health and wellbeing more holistically, has contributed to the delays in translating findings to practice.

3.4.3 Implications for exploring the health and caregiving experiences of fathers

The findings of this study reiterated many of the limitations of the systematic review on fathers in chapter 2, contributing to many of the design choices of the primary study on the health and caregiving experiences of fathers. Specifically, this study highlighted the need to consider fathers' positive experiences of caregiving alongside any challenges, as well as gaining a broader perspective of the types of support that may be beneficial to parents. It also raised important questions surrounding some of the assumptions of the caregiving literature; namely the impact of caregiving on health and wellbeing, and how this is usually framed and addressed. This must be balanced with the importance of understanding the issues that affect caregivers, though as discussed, our understanding of such is limited until the issues that underpin such outcomes, in the context of family caregiving, are considered. Not only did the maternal health study influence the design choices detailed in chapter 4, it also gave me the opportunity to gain a deeper understanding of the population in question, and helped me to develop my skills as a researcher at all stages of the process. This will be touched upon in more detail in chapter 6, both in the discussion surrounding the findings of the fathers study and in key lessons and reflections.

4. The health and caregiving experiences of fathers of children with a life-limiting condition: Methods

4.1 Introduction

Chapter 2 describes a systematic review and qualitative evidence synthesis that I conducted to explore the experiences of fathers of children with a life-limiting condition. Results highlighted some of the challenges experienced by fathers over the course of their child's illness including dealing with and adapting to uncertainty, role conflict, issues in the workplace, and not feeling recognised or listened to by healthcare professionals. The review also had several key limitations, which I aimed to address in the current study. Specifically, I aimed to address:

- The lack of information surrounding fathers as caregivers; the shift towards less dichotomous maternal and paternal roles has important implications in this context, particularly when we consider the family centred model of care in paediatric palliative care (WHO, 2023). Fathers have caregiving responsibilities, and therefore potential caregiver support needs. Although the review was able to identify the experiences of fathers spanning many years, across changing healthcare systems and medical developments, a contemporary study able to explore caregiving in the context of shifting parenting roles was warranted. The maternal health study provided an important benchmark for how the experiences of fathers might be explored, including how the roles of mothers and fathers may influences their experiences, health behaviours and interactions with services in the context of their own health.
- The lack of information surrounding the impact of fathers' experiences on their own physical and mental health, and therefore a lack of knowledge around how best to support these fathers. There is a body of research focused on the health of caregivers, but not specifically on fathers of children with a life-limiting condition.
- The overrepresentation of cancer studies, meaning that few studies addressed the population of children with complex neurodisability.
- The overrepresentation of US based studies.

4.2 Aims and objectives

My key research aims and objectives were:

- To describe the physical and mental health and well-being of fathers of children with a life-limiting condition using self-report measures.
- To describe the caregiving experiences of fathers both quantitatively and qualitatively.
- To describe fathers' demographic, family characteristics and health and well-being outcomes.
- To qualitatively explore fathers' perceptions of their own health and well-being and the factors that influence this.

4.3 Study design

4.3.1 Theoretical frameworks

The association between family caregiving and physical and psychological health is commonly conceptualised in relation to stress and coping (Lazarus and Folkman, 1984). Theoretical models demonstrate related factors that contribute to outcomes experienced by caregivers with many incorporating and building upon traditional models including the Stress Process Model (Pearlin et al., 1990), Appraisal (Lawton et al., 1991) and Risk-resilience (King et al., 1999). This is particularly relevant when we consider the issues raised in the review, such as shock, trauma, grief, coping and ongoing stress across many aspects of fathers' lives. Models also take other factors, such as socioeconomic factors, into account to highlight key contextual differences that must be accounted for in the examination of caregiver outcomes, and how such may mediate their experiences. This was particularly important to consider in the design of this study when we consider the heterogeneity of the population; not only in terms of a child's diagnosis and associated symptoms but the stage of their child's illness, current healthcare needs and prognosis. Furthermore, a broad model capable of capturing a wide range of factors was important when we consider the importance of context in this study i.e. to understand how fathers interpreted their experiences *within* their unique contexts.

Raina et al (2004), incorporate multi-dimensional constructs to the caregiving process and caregiving burden model, building on the work of Wallander et al. (1989), King et al. (1999)

and Pearlin et al. (1990) to create a combination of factors that can influence caregiver physical and psychological health outcomes (Raina et al., 2004). The model is based upon evidence surrounding the experiences of caregiving and affords the opportunity to explore positive dimensions of caregiving. The Informal Caregiving Integrative Model (Gérain and Zech, 2019) goes slightly further by incorporating aspects of occupational burnout with caregiver stress. This model defines caregiver burden as an individual's evaluation of the "physical, psychological, emotional, social, and/or financial consequences of caregiving" (Gérain and Zech, 2019), and it is this subjective burden that appears to be important in mediating the demands of caregiving with its outcomes, i.e. burnout. This further informed centralising this appraisal of caregiving in this study, and again allowed for the consideration of the positive experiences of caregiving as well as the equal consideration of determinants. Responses to difficult life events manifest uniquely within individual, social and familial contexts meaning that methodologies capable of exploring the implications of caregiving within a variety of contexts are required (Thin, 2018). The Informal Caregiving Integrative Model (Gérain and Zech, 2019) will be revisited in chapter 6, in which I will explore its relevance and detail how this thesis might be used to expand on/refine the model and shape further research. There was flexibility involved in using this model to frame the study; its use was exploratory, both in terms of its relevance and relationship between the factors.

4.3.2 A mixed-methods approach

Broadly speaking, the underlying assumption of mixed-methods approaches, i.e. the use of two or more methodological strategies in a single study, is that they have the ability to gain a more comprehensive understanding of a phenomenon than by using a single strategy (Bailey-Rodriguez, 2021), or specifically, quantitative or qualitative methodologies alone (Creswell and Clark, 2007). The literature surrounding the use of mixed methods research is vast. Nuanced, and often pragmatic, justifications for mixed methods approaches sit within individual studies, along with the understanding that rationale may emerge during the course of a study (Creswell and Clark, 2007). Greene et al (1989) defined some key rationale for combining quantitative and qualitative methods. First, and most often cited, is "triangulation"; used to obtain more valid conclusions through the comparison of both qualitative and quantitative results. Another rationale is that of "expansion"; the widening of enquiry to one of both sufficient breadth and depth i.e. through the use of closed ended survey questions and open ended interview questions (Creswell, 2003). Third, "complementarity" of methods allows for enhanced understanding of a phenomenon and greater opportunity for the identification of comparable and/or opposing views. It also allows for the description of general trends alongside elaboration of the details of such trends (Plano Clark, 2016). "Initiation" describes the possibility of new insights to help generate new research questions. Finally, "development" refers to using the results of one method as the foundations of the other (Schoonenboom and Johnson, 2017).

The rationale for using a mixed methods approach in this study was threefold. First, by using different methods, I was able to provide a general description of fathers' health issues which we know very little about compared to those of mothers, whose medical records are linked to their child's. Alongside this general description, the qualitative component was able to provide elaboration, explanation, and clarification of issues from fathers' perspectives, resulting in a more detailed understanding of their health and caregiving experiences and contextual factors that influence them. Second, the approach allowed for the opportunity to collect multiple perspectives and to identify and explore convergence and divergence between datasets i.e., triangulation. Third, the survey was able to act as a recruitment tool for the qualitative aspect of the study and provide flexibility in the way in which participants engaged with the study. This is referred to by Collins et al (2006) as part of "participant enrichment".

As the use of mixed methods research has proliferated across disciplines, though particularly in the health sciences, so have criticisms of its conceptualisation and development (Fàbregues et al., 2021). It is well supported as a methodology capable of assisting researchers to gain a more comprehensive understanding of a phenomenon, as discussed above. However, the incompatibility of the ontological and epistemological assumptions that underpin quantitative and qualitative approaches are central to methodological debate. Fundamentally, quantitative researchers emphasise a universal and objective reality whilst qualitative researchers support a socially constructed reality in which truth and meaning are "provisional, tentative and subject to change" (Dures et al., 2010). The underlying philosophical assumption underpinning mixed methods is *pragmatism,* in which knowledge is based upon experience and that the process of acquiring knowledge is a continuum, rather than part of the traditional philosophical dualism of objectivity and subjectivity (Denscombe, 2008). This disregards the unhelpful dichotomous divide between quantitative and qualitative methods, valuing both subjective and objective knowledge, grounding how we obtain knowledge in the questions to be answered.

Contemplating the rationale for mixed methods research, alongside its criticisms, are central to justifications for use. Such considerations are well documented in the literature, though are often prescriptive and without application to real-life or practical examples. Mason (2006) addressed these limitations through creative and accessible rationale, emphasising the need for multi-dimensional methods capable of capturing the complexity of human experience and relationships. The "heart and soul" of such can only be captured through multi-dimensional methods that "transcend or even subvert the so-called qualitative-quantitative divide" (Mason, 2006). If we consider this in terms of caregiving, we see that measuring the number of hours spent caregiving, or the number of caregivers experiencing mental health problems, does not tell us much about what else is taking place in unique individual, social, and familial contexts, in which relationships and roles are negotiated. Mixed-methods research allows us to include these multi-dimensional contextual factors, *how* they are relevant to the question, and how they may intersect (Mason, 2006).

This rationale does indeed lend itself to a "qualitative way of thinking" (Mason, 2006), and is the perspective taken in this thesis. The aim of the survey was to provide important descriptive information about fathers, though a qualitatively driven approach was warranted, given the need for more in-depth research, drawing upon fathers' own perspectives. Previous studies had touched upon fathers' emotional experiences, including feelings of shock, grief, trauma, sadness and worry (Chamberlain, 2007, Hayes and Savage, 2008, Wolff et al., 2010), though had not explored the impact of their experiences upon their health and wellbeing. Furthermore, few contemporary qualitative studies had sought to understand fathers' experiences through the lens of caregiving, meaning that scope for understanding dynamics within *family* caregiving was limited. Existing studies that sought to quantify levels of distress in parental caregivers (Vernon et al., 2017, van Warmerdam et al., 2019) were generally limited by maternal sampling biases and self-report measures that examine wellbeing through a pathological lens i.e., focused upon illness and distress. Such measures are useful in that they capture important information; often helping to identify appropriate means of support for those in need. However, they can often fail to account for positive experiences of caregiving and are often underpinned by the assumption that mental health and well-being are equivalent dimensions (Winefield et al., 2012). Furthermore, as mentioned in previous chapters, due to this focus on distress, they do not explore implications upon physical health nor fluctuations in health over time. Qualitatively driven data collection would provide a rich and detailed exploration and understanding of fathers' experiences, whilst helping to explain and elaborate upon the importance of selected variables and outcomes in the survey.

4.3.3 Epistemological position

The philosophical assumptions underpinning mixed methods, and the stance I took in the overall, initial design of this study is *pragmatism*. Indeed, a mixed methods approach was very much grounded in the aims and objectives of this study. The preceding sections provide the detail and rationale for this, fundamentally recognising the value of the continuum of subjective and objective knowledge. I also drew upon an interpretivist/constructivist paradigm to explore how fathers made sense of and gave meaning to their experiences, whilst recognising the active role of my own experiences in interpreting such data (Creswell and Creswell, 2017). There is a recognition within interpretivism that patterns of meaning arise as a result of interpretation, reproduction or construction, rather than as the result of an absolute and external truth, whilst the interpretivist stance emphasises the importance of context over the desire for generalisable findings (Schwandt, 1994). The aim here was to understand how fathers interpret their experiences within their unique contexts; to explore which aspects of such contexts impacted their health and wellbeing outcomes and recognising value in depth and nuance. Reflexive thematic analysis is particularly compatible with this stance, a method for which the researcher plays an active, interpretive role…"themes do not passively emerge from data" (Braun and Clarke, 2019). Critics of qualitative methodologies, and associated epistemologies, emphasise these 'challenges' in generalisability, despite it not being a common goal of such methods (Leung, 2015). However, although this desire for generalisability is not a priority, collecting diverse experience through qualitative methods and expressing that data in the form of analytical themes does allow for an understanding of *shared experience* and transferability (Maxwell, 2021).

4.3.4 Timing and integration of quantitative and qualitative components

Approaches to the integration of quantitative and qualitative methodologies are laid out in mixed methods guidelines, which define various levels of integration based on the research aim and specific mixed methods design. There have been numerous delineations of mixed methods typologies (Schoonenboom and Johnson, 2017, Teddlie and Tashakkori, 2009, Leech and Onwuegbuzie, 2009, Johnson and Christensen, 2014) but perhaps the most widely used are those identified by Creswell and Plano Clark (2011). The authors define six core designs; (1) convergent, in which the implementation of qualitative and quantitative strands occurs concurrently, the strands of which are analysed separately and merged during interpretation and analysis; (2) explanatory sequential in which qualitative results help to explain the results of quantitative data; (3) exploratory sequential in which the qualitative strand precedes the quantitative strand, the purpose of which is to test qualitative findings; (4) an embedded design in which one strand is added to the other for the purpose of support. Usually there is the prioritisation of one strand and data collection can be concurrent or sequential; (5) a transformative design in which a transformative theoretical framework provides to foundations of the study; and (6) multiphase involves a combination of concurrent and/or sequential data collection over more than 2 phases. A key decision in this study related to the offer of an interview at the end of the survey, meaning that at a participant level, the quantitative element would come first. However, at a study level the quantitative and qualitative data were collected and analysed during a similar timeframe, i.e., a convergent design (Creswell and Clark, 2007, Creswell and Plano Clark, 2011). It is important to add that within the convergent design, I used an interactive approach, in that initial findings from the survey informed some of the questions asked at interview i.e. integration at a methods level. Furthermore, initial qualitative findings also shaped subsequent interviews demonstrating features of some of the other typologies noted above.

A key concern in any mixed-methods approach is how and when each methodological strand should be integrated. Approaches to the integration of qualitative and quantitative elements can be implemented at any level of the research process including research question, theoretical drive, design, methods, analysis, interpretation, and reporting (Fetters et al., 2013). However, understanding the design and rationale for combining qualitative and quantitative approaches does not provide much insight into the practicalities of integration i.e., *how* findings can be combined in a meaningful way. The extent to which existing studies integrate their findings does vary, as do definitions of integration (Creswell and Plano Clark, 2011, Creswell and Creswell, 2017, Fàbregues et al., 2020). A general understanding surrounds the ways in which qualitative and quantitative components are connected, which can be at any level of the research process including research question, theoretical drive, design, methods, analysis, interpretation, and reporting. Critique of mixed methods approaches often focuses on how well findings are integrated during the latter stages of a study i.e., have the results been integrated with one another or rather summarised alongside one another? (Bryman, 2007). More recently, novel methods of integration have been reported, allowing for conclusions beyond those of the individual threads. Narrative, transformation, or joint display approaches have all been suggested as means of integration at interpretation and reporting stages (Fetters et al., 2013). Narrative approaches present the findings of each thread separately, whilst providing a written account of the relationship between them. Transformation involves integration through transforming one dataset into the same type as the other whilst joint displays offers opportunities for the discovery of new insights through the side-by-side presentation of data (Fetters et al., 2013)

In this study, integration occurred at multiple points. In its broadest sense, it occurred during the design of the study through deciding which objectives would be met through each element of the study. The elements were also linked through the caregiving frameworks 4.3.1 from which I established that key contributors could not be adequately captured through quantitative or qualitative data collection alone. Integration also occurred as participants were recruited to the interview via the survey and emerging data from the survey was used as prompts in the interviews. At an interpretation and reporting level, the data were integrated through a narrative joint display table of the findings from each component (chapter 5) (Fetters et al., 2013, Skamagki et al., 2024). A summary of the study design can be seen in figure 5 below. In chapter 5, I first present the findings of the quantitative and qualitative components separately. I then include a joint-display table in which the findings of each thread are presented side-by-side (Skamagki et al., 2024). Through this table, links between the data were established including similarities and differences; meta-inferences are presented as a new column in the table. As this study comes mainly from a qualitative perspective, the qualitative themes, and sub-themes, were used as the overarching concepts in the joint display table. I then mapped the findings of the survey onto these themes/sub-themes. Interpretations are displayed in the final column of the table and are further discussed in chapter 6.

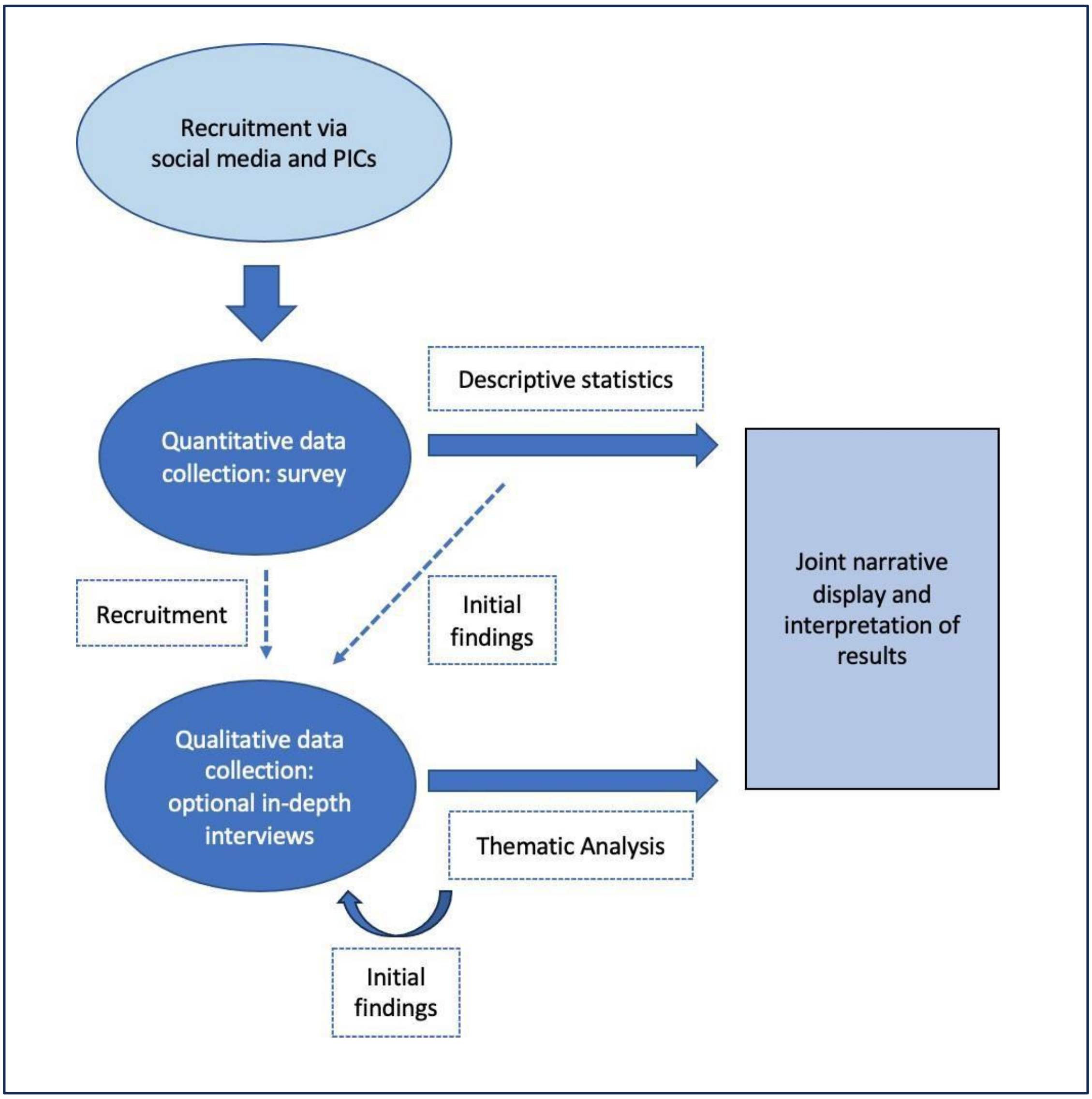


Figure 5; Summary of research design

4.3.5 Quantitative component

4.3.5.1 Setting

I recruited fathers of children with a life-limiting condition from three children's hospices and one NHS children's hospital in England.

4.3.5.2 Sampling

Purposive sampling was used to recruit fathers of children with a life-limiting condition. This included fathers of children with a range of diagnoses and from a range of backgrounds. It included fathers with varying degrees of caregiving responsibilities.

The inclusion criteria required that participants:

- were aged 18 or above;
- were the father of a child with a life-limiting condition;
- had the capacity to consent to the study.

Fathers were excluded from the study if their child had died.

'Father' included stepfathers, adoptive fathers, long-term foster fathers and biological fathers. A life-limiting condition was defined as one falling into one of the four Together for Short Lives categories as shown in chapter 1 (TfSL, 2018).

4.3.5.3 Initial recruitment to survey

The aims of the survey (Appendix 7) were to; collect descriptive information about fathers including demographics: health and well-being; sleep; caregiving and to explore demographics and family characteristics as well as health and well-being outcomes. The initial target sample size for the survey was 150 participants. Questions were tested with members of the PPI panel to assess readability, length, and content of the survey. The PPI members felt that some slight amendments to the introductory page were needed, in terms of general re-wording and to make the inclusion criteria clearer.

Several approaches were used to identify and recruit participants, as not all children diagnosed with a life-limiting condition are supported by children's palliative care services (e.g. children's hospices, paediatric palliative care teams). Furthermore, fathers are typically a hard-to-reach population and so flexible recruitment strategies were necessary. Fathers were able to take part in the survey **or** survey plus an interview. I used two recruitment routes:

4.3.5.3.1 Hospices/ NHS Trusts

Clinical staff informed fathers of the study in face-to-face consultations, over the phone, via email, or via post, depending on their usual means of contact with parents. They provided fathers with an invitation letter (Appendix 8) and information sheet (Appendix 10) detailing the purpose of the study (including details about the quantitative and qualitative components). Once fathers had all of the information, they were directed to the survey URL landing page if they want to take part. The landing page for fathers referred by hospices/NHS trusts contained the electronic consent process with the option of reading the information sheet again if needed. However, they were provided with the information sheet by a healthcare professional and so it was not mandatory to read it again online before they start the survey.

4.3.5.3.2 Social media advert

The study was advertised on social media (Twitter) (Appendix 9) with a short summary of the project and a link to the landing page of the survey (and information sheet and electronic consent procedure).

4.3.5.4 Survey data collection

I collected data via the survey between November 2021 and March 2023. This was such a long recruitment window as I took maternity leave during this period. The survey was open and had been advertised through social media, but no active recruitment was taking place through PICs. Active recruitment via PIC's started in July 2022. The self-report electronic questionnaire was administered via Qualtrics (Qualtrics, Provo, UT) and collected demographic information about fathers, their child, family structure, sleep, caregiving and information regarding their own physical and mental health and well-being. Fathers were also able to complete the survey over the phone with a researcher if they are unable to complete it electronically. If they wished to do so they could contact a researcher to arrange a time and date.

4.3.5.4.1 Background and context

Relevant participant demographics were self-reported, including age, marital status, family structure, education, employment, household income, ethnic origin, geography (region). Questions were selected from national surveys (Statistics, 2021, Government, 2018), and were based upon key variables identified in the theoretical framework (Raina et al., 2004, Gérain and Zech, 2019), were capable of socially locating fathers' experiences. Fathers answered questions regarding their caregiving responsibilities; whether they were their child's primary or secondary caregiver, or whether they had shared caregiving responsibilities with their partner. However, it is important to note that there were no constraints under which fathers described their caregiving roles i.e. I did not include definitions of 'primary' and 'secondary' caregiver.

4.3.5.4.2 Child characteristics

Information regarding the child's diagnosis, functioning (question selected from Collins et al., 2020 (Collins, 2020)), care needs, sex, age, age at diagnosis and any hospice support were captured.

4.3.5.4.3 Appraisal of caring

The 25-item Family Appraisal of Caregiving Questionnaire for Palliative Care (FACQ-PC) (Cooper et al., 2006) was used to asses fathers' experiences of caring for a child with a life-limiting condition. Four sub-scales made up the scale; caregiver strain; caregiver distress; positive appraisal of caregiving; family well-being. A higher mean score (between 1 and 5) for each sub-scale indicates a greater level of the issue being assessed within each sub-scale. The scale has an internal reliability of 0.75 to 0.86 (Cooper et al., 2006). Not only would this scale allow for the exploration of positive experiences of caregiving, an important part of the study, but it would allow me to compare fathers' appraisals to those of other parental caregivers in other studies (Collins, 2020).

4.3.5.4.4 Physical and mental health state

Questions relating to general health and whether participants have any long-lasting health conditions or illnesses that have lasted or expected to last for 12 months or more. This captured information relating to fathers and will include a list of common physical and mental health diagnoses. I selected questions based on those used in national surveys (Statistics, 2021, Government, 2018).

4.3.5.4.5 Sleep

Participants completed the PROMIS Sleep Disturbance- Short Form 8a (Hanish et al., 2017). This 8-item measure asks participants to rate their sleep quality between 5 (very poor) and 1 (very good) and to respond to statements identifying any sleep issues (e.g. difficulty falling asleep). Each item is scored between 1 and 5. A higher score indicates greater sleep disturbance. Major sleep disturbances are cited in the literature on parental caregiving (Kim and Rose, 2011, McCann et al., 2014) and has important implications for physical and mental health and wellbeing (Gallagher et al., 2010). The inclusion of this scale would allow for comparisons with average scores.

4.3.5.4.6 Health-related quality of life (HRQoL)

The EQ-5D-5L allows for participants to subjectively describe and value their own HRQoL. It is made up of 5 dimensions; mobility; self-care; usual activities; pain/discomfort; anxiety/ depression and each of the dimensions has 5 levels; no problems; slight problems; moderate problems; severe problems; extreme problems. Participants indicated their health state by selecting the most appropriate statement for each dimension. 1-digit scores for each dimension were then combined into a 5-digit health state. Health states were then converted into a single index value using a UK specific value set (EuroQol). The EQ-5D also contains a visual analogue scale through which participants rated their health between 100 'the best health you can imagine' and 0 'the worst health you can imagine' (Herdman et al., 2011).

4.3.5.5 Survey data analysis

As mentioned above, the initial target sample size was 150 participants and would have allowed for slightly more sophisticated analysis of the survey data. I had initially planned to use χ2 and independent t-tests as appropriate to investigate the association between health outcome variables and sociodemographic/family characteristics. I was then planning to explore variables thought to impact upon outcomes using multivariable logistic regression analysis. However, the final sample size did not allow for this type of analysis. Not only would this have provided more insight into health outcomes in this population, but it would also have impacted the way in which the quantitative and qualitative strands were integrated following analysis which I will touch upon below in section 4.3.7.

Basic descriptive statistics were used to describe the characteristics of participants and their children, health conditions, the distribution of scores from EQ-5D-5L and caregiving scales. Initially, it was envisaged that I would investigate the association between health outcomes and sociodemographic characteristics/caregiving experiences. However, the final sample size did not allow for this, and so descriptive statistics were used to summarise the characteristics of the sample. Furthermore, the data were integrated with the qualitative data through interpretation and reporting (Fetters et al., 2013), which is outlined further below.

4.3.6 Qualitative component

4.3.6.1 Recruitment to interview

Fathers were made aware of the qualitative aspect of the study when they are invited to take part in the survey by healthcare professionals and on social media. Upon completion of the survey, participants were able to submit their details for me to contact them about the qualitative aspect of the study. I then contacted them to discuss the study in more detail, provide them with an information sheet that referred to the qualitative interview only and organise an interview if appropriate.

4.3.6.2 Qualitative sample size

I aimed to recruit between 25-30 participants. This was a provisional and flexible target based upon the number of participants required to generate rich and meaningful data as well as what was considered to be feasible in the given timeframe (Braun and Clarke, 2021b).

4.3.6.3 Qualitative data collection

I collected data between April 2022 and March 2023. As recruitment was active whilst I was maternity leave, one of my colleagues (a senior research fellow with experience in qualitative interviewing), conducted the first two interviews with fathers. Semi-structured interviews were conducted on Zoom or over the telephone, depending on participant preference. Face-to-face interviews were not possible due to ongoing Covid-19 restrictions. With use of a topic guide (Appendix 11), interviews explored fathers' experiences of being a father of a child with a life-limiting condition, addressing their caregiving roles, perceptions of coping and sources of both formal and informal support, their relationship with healthcare professionals and any occupational role conflicts. Fathers' accounts of their own physical and mental health were explored. The topic guide was developed using concepts from the multi-dimensional model of caregiving (Raina et al., 2004) and the Informal Caregiving Integrative Model (Gérain and Zech, 2019) and with the input of the Family Advisory Board (see PPI section below). The topic guide was used to ensure that the interviews were consistent and addressed the key aims of the study but with the flexibility for fathers to talk about their own unique experiences and perspectives of things that were important to them in the context of their own health. This also allowed for comparisons between individuals (Flick, 2017).

4.3.5.9 Thematic analysis of interview data

Qualitative data was analysed using Braun and Clarke's reflexive thematic analysis (Braun and Clarke, 2021a, Braun and Clarke, 2021b, Braun and Clarke, 2019, Braun and Clarke, 2024). In identifying shared patterns of meaning across the dataset, I developed themes surrounding the health and caregiving experiences of fathers. This approach is centralised upon the active role of the researcher, aligning with my own constructivist/interpretivist positioning described earlier in this chapter. Despite using the theoretical frameworks to guide the design of this study, analysis was inductive. There were six phases to the process of reflexive thematic analysis, as defined by Braun and Clarke (Braun and Clarke, 2006, Braun and Clarke, 2019). However, this process is iterative and flexible, rather than linear and has been defined as a guide, rather than a strict analytical procedure.

(1) *Familiarisation*: I made notes throughout each interview and afterwards spent some time reflecting upon and making notes about the interview. I debriefed with a colleague after each interview. The interviews were audio-recorded and transcribed verbatim. A professional transcription service was used for transcribing the interviews. Analysis would usually begin during transcription which is "inherently interpretive" and "influenced by the transcriber's own assumptions and biases" (McMullin, 2021). However, to minimise the impact of using a transcription service, I reviewed each transcription against its audio recording, and then listened again whilst making notes. This helped me to immerse myself in the data, mitigate against any transcription errors and capture nuance and context in the data (Byrne, 2022).

(2) *Systematic data coding:* NVivo was used for managing and coding data. Analysis was inductive, in keeping with the exploratory nature of the study. I worked through each transcript one-by-one, coding data line-by-line using a mixture of semantic and latent coding. For example *"I'm mentally exhausted*" was coded as 'mental exhaustion' i.e. semantic coding. Latent coding encompassed a greater level of interpretation, for example, "*The best she could come up with was some deep breathing exercises. The following week she was telling me about therapeutic doodling"* was coded under 'lack of appropriate psychological support'. Data could be coded more than once and could be coded both semantically and latently if necessary. This was an iterative process during which I named and renamed codes as necessary. I frequently discussed the coding process and initial codes and themes with the wider research team.

(3) *Generating themes:* Once all data had been coded, I examined the relationship between the codes to find *shared meaning* and initial themes. Again, these were discussed with members of the wider team and with members of the family advisory board for their reflections. An important feature of this type of thematic analysis is that themes are developed from shared meaning of a concept rather than a shared topic. Generally, themes should go beyond topic summaries or categories, often with the theme name being illustrative of a story in itself (Braun and Clarke, 2024). For example, the first theme in this thematic analysis revolved around fathers' busy schedules, with vivid descriptions of their caregiving routines and daily responsibilities. Initially, codes were grouped together on this basis, and through further engagement and reflection, a theme was generated around the fragility and unpredictability of fathers' daily routines.

(4) *Reviewing potential themes:* I reviewed potential themes in relation to coded data items and to the dataset as a whole. Stages 3, 4, and 5 were particularly iterative, with boundaries between them hard to distinguish. Themes were developed through deep and reflexive engagement with the data that evolved throughout the process rather than as a result of a checklist of procedures or "coding for themes" (Braun and Clarke, 2024). The re-reading of transcripts and of my notes taken during the interviews and analysis, alongside assessing the 'story' that was being told by the initial themes, the

reflections of PPI members and colleagues, allowed for iterations that strengthened and elaborated upon these early themes. There were many aspects of fathers' experiences that influenced this sense of fragility mentioned above, many of which also impacted their ability to maintain or establish relationships with others. Therefore, an initial theme surrounding relationship related issues was incorporated into this first theme. The shared meaning across each of these concepts was the uncertainty, fragility, and inflexibility that fathers' experienced in relation to, or as a result of providing care for their child.

(5) *Defining and naming themes:* Again, the generation and naming of themes is a subjective process, requiring immersion in and an understanding of the data. It is not a case of simply discovering themes that happen to naturally exist within the data nor is it a case of finding 'answers' or 'truths'. It rests upon this immersion and exploration. There are multiple iterations of what we consider to be 'thematic analyses' but in recognising, accepting, and encouraging the inclusion of the researcher as an active part of this process, we are able to gather important and unique interpretations of data.

(6) *Producing the report:* Data-driven themes were reported after the findings of the quantitative survey, followed by a joint display of findings as discussed above. Themes were assessed and discussed in relation to the caregiving literature in chapter 6.

The extent to which this was *reflexive thematic analysis,* as opposed to other general forms of thematic analysis (Braun and Clarke, 2019, Braun and Clarke, 2024), rests on the acknowledgement of my role in knowledge production. Ongoing reflexive engagement with my own beliefs, background, and assumptions, my own development as a researcher, and how each of these influenced data analysis was key in the analytical work required (Braun and Clarke, 2019).

4.3.7 Integration

As mentioned above, the quantitative and qualitative threads were integrated at various points throughout the research process. In terms of integration at the interpretation and write-up stage, findings were presented in a narrative joint display table (Skamagki et al., 2024). To recap, this is a visual display of key findings from each of the threads, which were in this case presented with respect to each of the qualitative themes and sub-themes. Once I had created the table, I searched for links between the qualitative and quantitative components and mapped relevant quantitative data onto the qualitative themes. Exemplar quotes were also used to represent the qualitative themes/sub-themes. This enabled the data to be synthesised together, drawing out key findings and a deeper exploration of the data, whether that be through areas of convergence, divergence, expansion, or complementarity (Skamagki et al., 2024).

In terms of the plan for integration, the stage at which the findings were integrated was always going to be at the interpretation stage i.e. following separate analyses. However, the level of explanation achieved through integration may have been greater if I had been able to carry out the quantitative analyses as originally planned, looking at various outcomes between demographic groups etc. As a qualitatively driven study, the level of important, rich, and meaningful data was enough to make some key recommendations for research and practice (see discussion). Though I think it is important to note here that I did initially plan to compare, contrast, and explain the quantitative data to a greater extent than was possible. This is more of a discussion point, though also has implications for recruitment strategies and questions surrounding *how* we might collect important data on the health outcomes of fathers.

4.3.8 Ethical considerations

Ethical approval was obtained for the study from the London- Bloomsbury Research Ethics Committee (REC reference 21/LO/0591- see Appendix 11). This process was challenging as initial comments from the REC suggested that I should take consent from the child, if fathers wanted to discuss their caregiving experiences with me. The ethical issues associated with qualitative research are vast with guidance to assist researchers in upholding sound ethical principles (Taquette and Borges da Matta Souza, 2022). However, how researchers navigate the ethical challenges specific to paediatric palliative care research has not been widely addressed meaning that there is little guidance for researchers, HCP's and for research ethics committees (REC's) in this context. This presents additional barriers to conducting research with an already underrepresented population. The considerations surrounding caregivers in adult palliative care research offer some closely related guidance (Abernethy et al., 2014), though are not entirely transferable when we consider the nuanced dynamics of a paediatric setting as well as the diversity of the population in question. To satisfy the REC, I amended the topic guide for the interviews to make it clear that my intention was not to collect any identifiable information about a child or young person, and that any such information collected as part of fathers' accounts would be redacted prior to analysis, as is the case in most interview studies. I also made it clear that collecting relevant information as part of the survey, i.e., child's age and health, would help me to build context around fathers' own health and experience but were not identifiable in nature. Although ethical approval was granted, without the requirement of taking consent from the child, this highlights the need for further research and guidelines to outline key ethical issues and means of addressing them. Central to this is that individuals should not have to seek permission from others to be able to share their own experiences and perspectives.

Names and other identifying information, such as specific hospital or hospice names, were redacted from the interview transcripts. Ethical concerns remained surrounding the identifiability of participants given the rarity of some of the conditions included in the study. Therefore, specific conditions were also redacted. Participant characteristics were provided as group variable summaries.

As mentioned above, potential participants were informed about the study via social media or were approached by staff at PIC’s. Recruitment via social media is increasing (Gelinas et al., 2017), allowing for advertisement to larger and more diverse samples (Darko et al., 2022). Critique of such methods of recruitment mainly surrounds aspects of privacy and confidentiality issues, as well as concerns surrounding those who cannot access social media. In this study, social media recruitment, via X (formerly Twitter), was used to supplement recruitment at PICs, and participants were signposted to the Qualtrics survey. Although I handled all information appropriately and in line with data protection guidelines i.e., through appropriate storage and the removal of any potentially identifiable information, I recognise that debates surrounding social media recruitment are ongoing and complex. Third-party tracking of internet activity has been raised as a key concern in the online recruitment of participants to health studies, though it is recognised that such risks need to be balanced with the benefits of widely accessible and cost-reducing recruitment methods (Bender et al., 2017).

Potential participants (regardless of recruitment route) were given the full information sheet for the study (quantitative and qualitative components) before taking part in the survey. They provided consent prior to taking part in the survey, and if they decided to take part in interview as well, they were given another information sheet detailing the interview process further. All participants gave written or verbal consent prior to the interview and were given the opportunity to ask any questions about the interview and time to consider participation. I made it clear to participants that they could stop the interview or withdraw at any time, without giving reason. I also explained that the interview was confidential, unless they told me something that caused concern in relation to their own, or someone else’s, safety. I explained that I would discuss this with them if that happened.

If participants became distressed during the interview I asked if them if they would like to stop, take a break or rearrange it. Identifying and addressing participant distress is key to ensuring the ethical nature of interviews (Whitney and Evered, 2022), though taking a break, stopping, or rearranging the interview should generally be decided by the participant. Distress protocols that are able to address potential and definite distress are helpful in this respect, as they avoid reliance upon the interviewers judgement and own emotional projections (Whitney and Evered, 2022). Some fathers did become tearful during the interviews, but all wanted to carry on. Research suggests that in-depth interviews can have a positive emotional impact on interviewees, offering them a chance to offload. Equally, researchers need to be aware of the potential for participants to experience unexpected distress or traumatisation through talking about their experiences (Pascoe Leahy, 2021). At the end of each interview, I asked fathers if they consented to me contacting them in the 24-48 hours following the interview. This gave them the opportunity to consider whether they wanted to add to or clarify anything in their accounts, as well as for me to ask how they felt. This is in line with recommendations surrounding follow up (Pascoe Leahy, 2021).

Another key part of upholding an ethically sound process, was through reflexivity i.e., a critical analysis of my own influence on the research process (Taquette and Borges da Matta Souza, 2022). It is important to note here alongside other ethical considerations as "responding to ethical dilemmas arising 'in the moment' requires a reflexive approach whereby the researcher questions his/her own motivations, assumptions and interests" (Reid et al., 2018).

4.4 Patient and Participant Involvement

The family advisory board (PPI panel introduced in chapter 1) contributed to the decision to use a mixed methods approach of this study. Below, I detail their specific contributions to this study:

- The group helped me to make key decisions surrounding methods of data collection, including the decision to employ a mixed methods design with an optional interview. This would improve flexibility and allow fathers the decision as to whether they wanted to discuss their experiences with me. All of the fathers in the group felt that although the interviews were valuable, many fathers may want to share their experiences outside of interactions with a researcher. They helped me with the wording and format of the survey questions, particularly the wording of the eligibility criteria and introduction to the study. Fathers were keen for me to make the benefits associated with the study clear.
- The group piloted my survey and gave me feedback on ease of completion, content, and length. Fathers felt that the survey was acceptable in length and recommended the addition of a progress bar.
- The members reviewed my initial themes; these themes resonated with fathers (and mothers) on the panel and discussions helped to further clarify themes. These are similar to member reflections (Tracy, 2010), through which I was not looking for accuracy checks, but for further perspectives and elaboration. This led to discussions surrounding the implications of these findings, particularly around the type of support needed by families. This is discussed in chapter 6.

4.5 Reflexivity

Reflexivity, and reflexive thematic analysis as explored above, encourage researchers to recognise and accept how their own beliefs, position and assumptions play a role in influencing all phases of research from development to the interpretations of data (Campbell et al., 2021). During this study, there were a number of ways that I felt my own positioning may have impacted the research process and findings. I also experienced several challenges during the research process, that prompted me to further consider my role. First, in interviewing participants, I formed social relationships with them. I built rapport with participants prior to the interviews, over email or the telephone. I used these opportunities to introduce myself, find out a little more about the participant, and answer any questions they had. Additionally, it gave me the chance to assess the pace and tone of questioning that might be appropriate for individual participants. Although this was an ongoing, evaluative process throughout each of the interviews, instances in which I was able to speak to the participant beforehand helped enormously in my own preparations, not only with such issues of pace and so on, but in helping to ease any apprehension I had. My apprehension was mainly related to wanting to get the best from the interview; to put the participant at ease and to create a space in which they felt comfortable to share their experiences. The participants were extremely forthcoming about their experiences, and many had been pleased to see that there was a study focused specifically on fathers. I started the interviews by asking generally about themselves and their families. Most fathers talked about their child/ren, including their age/s, condition and needs. I then moved onto questions surrounding fathers' own health and wellbeing. In chapter 3, I presented the maternal health study. In those interviews, I approached questioning in a similar way i.e., asking about the child, followed by questions about mothers' health. I sometimes found this transition to be uncomfortable, related perhaps to the pace at which I shifted my line of questioning, and mothers' responses to such. It could feel like I had restricted the conversation, changed the pace, or focus, at the wrong moment, and it took time to recalibrate the conversation. Upon reflection, I had felt pressured to move onto questions directly related to the health of mothers, in line with the specific aims of the study and struggled to do this without explicitly moving the interview on. This may have seemed abrupt to mothers, who had often given me a lot of information about their child. There was not a problem with moving the interview on *per se*, but I felt I had missed out on opportunities to thank mothers for sharing information about their child and that this information was really valued. This was heavily linked to my skills and confidence in conducting interviews and is just one example of how post-interview reflections led to changes in the way I approached them.

The interviews with fathers felt more comfortable. I was less concerned with covering the points of the topic guide in a prescriptive manner and had developed the skills and confidence to gently direct the conversation whilst ensuring that we were covering topics as per the aims of the study. In discussing incredibly sensitive and emotional topics with these fathers, it could become distressing to hear about challenges they were currently facing whilst knowing that there was nothing I could do to help, despite knowing that it was not in my role to do so. This was linked to the frustration I felt in hearing similar stories repeated, surrounding examples of fathers' perspectives of being neglected by services, especially during the COVID-19 pandemic. It helped me to discuss these frustrations during debriefs to ensure that I did not carry them into subsequent interviews.

In terms of my own background and positioning, there were several factors that may affected my research. First, prior to undertaking this PhD, I completed a master's degree in psychology. I had developed key interests in parental mental health and held several assumptions about the benefits of psychological interventions and their potential for use with this population. I wonder how this manifested during those early interviews with mothers when I asked about any specific support that they needed. Second, and most notable to me, is that I became a mother during the study. Although my child is healthy, my perspectives of parental roles did change significantly, and I wondered if this would affect the extent to which I resonated with stories of parenthood. Furthermore, I wondered if this would lead to a prioritisation of these stories that I resonated with most, which I know can be a common problem in qualitative data analysis. I spent a lot of time reflecting on my thoughts and feelings following each interview, particularly thinking about how my views on and connection with individual participants may have influenced the analysis. This, coupled with debriefs with other team members, helped me to maintain the capacity to attend to numerous perspectives.

As a woman, I was concerned that some fathers would be reluctant to share their experiences with me. I wondered if I would be perceived as aligning more with the perspectives of mothers, particularly during interviews in which fathers talked about challenging relationships with their partner. This was sometimes brought into focus when participants would ask questions such as, "have you got children?". Self-disclosure is something I had naively not thought about prior to conducting the interviews, and at first found it difficult to respond to such questions, often aiming to deflect them. However, research does advocate for a certain level of interviewer self-disclosure, particularly in semi-structured approaches, as a means of building rapport and breaking down any perceived power imbalances (Abell et al., 2006). As I gained more experience in interviewing, I became more comfortable with sharing small pieces of information about myself with participants. This did not happen frequently, but in answering participants' questions without deflection, the flow of conversation was maintained. Fathers were generally very open about their experiences with me. I wonder if any perceived distance from their experiences may have facilitated this openness, as opposed to a potential lack of clarification of their experiences had they thought we had more in common.

4.6 Conclusion

In this chapter I have set out a detailed account of the mixed methods study design. I have detailed my methodological approach and rationale, emphasising the need for an exploratory and flexible study design; a quantitative interview followed by an optional qualitative interview. I have outlined the value of qualitatively driven mixed-methods research, emphasising the role of reflexive and interpretive engagement throughout the research process. I have considered my own positionality, particularly in how I collected and analysed the interview data, recognising how this may have shaped the findings. In chapter 5, I go on to present the findings of the study.

5. The health and caregiving experiences of fathers of children with a life-limiting condition: Findings

This chapter presents the findings of the online quantitative survey and the semi-structured qualitative interviews. I start by summarising the survey findings which includes descriptions of fathers' characteristics and demographics, health, sleep, and caregiving appraisals. As outlined in chapter 3, the aim of the survey was to provide some key contextual information, socially locating fathers' experiences whilst also acting as a recruitment tool for the interviews, allowing fathers to decide whether they wanted to share their experiences in more depth. Following on from the tabular summaries of each part of the survey, I will present the findings of the thematic analysis of the interview data; 12 of the fathers that completed the survey went on to take part in an interview. Quantitative and qualitative data will finally be presented in a joint display table, highlighting key findings which will be further explored in chapter 6.

5.1 Survey findings

A total of 32 individuals completed the survey between March 2022 and March 2023. These participants completed the entire survey and were included in the final results. 12 further individuals started but did not complete the survey and were classed as having withdrawn from the study as per the participant information sheet and ethics approval.

The age range of the 32 participants was 36-54 years. Fathers from across the UK took part. The ethnicity of most of the fathers was White (94%). The 2 participants from ethnic minority groups are reported together as categories were so small as to potentially identify participants (Flanagin et al., 2021). Most of the fathers were born in the UK (94%). The majority described themselves as either their child's primary caregiver (n=16) or as having shared caregiving responsibilities with their partner (n=9). The remainder of the fathers (n=7) described their child's other parent as being their primary caregiver. In terms of employment, 15 fathers were in full-time employment, 12 had home/caring responsibilities, and 5 worked part-time. 11 fathers had stopped work to care for their child, 9 had reduced their hours and 6 fathers had made changes to their career path to care for their child. The mean time since their child's diagnosis was 10 years. Most fathers lived with their child's other parent. Table 2 summarises participant characteristics and demographics.

Table 6; Fathers' characteristics and demographics

Participant characteristics (n=32)	
Age (range)	**26-54 years**
Ethnic group	**n (%)**
English/Welsh/Scottish/Northern Irish/ British	30 (94)
Other ethnic minority group	2 (6)
Location in the UK	**n (%)**
East of England	5 (16)
East Midlands	10 (31)
North-East	2 (6)
London	9 (28)
Yorkshire and the Humber	4 (13)
South West	2 (6)
Religion	**n (%)**
Christian	11 (34)
Jewish	1 (3)
Muslim	2 (6)
No religion	18 (56)
Highest qualification	**n (%)**
GCSE	6 (19)
A-Level	7 (22)
Bachelor's degree	11 (34)
Master's degree/ PhD	7 (22)
Vocational qualification	1 (3)
Employment status	**n (%)**
Home/ caring duties	12 (37)
Full-time work	15 (47)
Part-time or casual work	5 (16)

Participant characteristics (n=32)	
Employment change following child's diagnosis	**n (%)**
Stopped work to care for child	11 (34)

Reduced hours to care for child	9 (28)
Change in career path	6 (19)
No change in career	6 (19)
Caregiving role	**n (%)**
Child's primary caregiver	16 (50)
Joint caregiving responsibilities	9 (28)
Child's other parent is their primary caregiver	7 (22)
Household income per year before tax	**n (%)**
<£10,000	1 (3)
£10,000-24,999	11 (34)
£25,000-49,000	6 (19)
£50,000-74,999	4 (13)
£75,000-99,000	7 (22)
£100,000 or more	3 (9)
Relationship	**n (%)**
Married or domestic partner	29 (91)
Separated	2 (6)
Single, never married	1 (3)
Number of children	**n (%)**
1	12 (37)
2	13 (41)
3+	7 (22)

32 fathers of 38 children, aged 3-23, took part in the survey. The individual diagnoses of the children varied. Most of them had been diagnosed with a neurological (n=12) or genetic (n=17) condition, and most had multiple diagnoses i.e. medical complexity (n=26). The mean age of diagnosis was 11.7 years and 34 children had diagnoses that meant they required significantly more care, most or all of the time, compared with children of a similar age. There

was a mix of children who did and did not receive care from a children's hospice. Table 3 summarises the characteristics of the children.

Table 7; Child characteristics

Child characteristics (n=38)	
Life-limiting condition diagnostic category	n (%)
Cardiac	3 (7.9)
Congenital	4 (10.5)
Genetic	16 (42.1)
Metabolic	1 (2.6)
Neurological	12 (31.6)
Unknown or no formal diagnosis	2 (5.3)
Age	
Range (years)	3-23
Mean age (years)	11.7
Sex	n (%)
Male	18 (47.4)
Female	20 (52.6)
Age at diagnosis	n (%)
Before birth	2 (5.3)
At birth	4 (10.5)
Infancy (0-1 years)	9 (23.7)
Childhood (1-9 years)	21 (55.3)
Unknown	2 (5.3)
Needs relative to other children of similar age	n (%)
Significantly more care; most or all of the time	34 (89.5)
Moderately more care most of the time	4 (10.5)
Children's hospice user	n (%)
Yes	28 (73.7)
No	10 (26.3)

Fathers described some health problems, including mental health problems, back problems, arthritis, and high blood pressure. No other specific health concerns were reported in the survey. Raw summary scores from the PROMIS sleep disturbance scale were used to describe general sleep disturbance (Yu et al., 2012). Each item in the scale is rated on a 5-point scale, meaning that total raw scores can range between 8 and 40, with a higher score indicating higher sleep disturbance. These scores were converted to a standardised T-score (with a mean of 50 and SD of 10) based on recommended scoring. Participant T-scores ranged from 38.1 to 77.5.

Table 8; Fathers' health and caregiving appraisals

Question or measure		
Physical or mental health concerns	n (%)	
Yes	18 (56.3)	
No	14 (43.7)	
Nature of concern reported	n (%)	
Mental health problems	13 (40.6)	
Arthritis	2 (6.3)	
High blood pressure	4 (12.6)	
Back problems	5 (16.6)	
Other illness or progressive disability	1 (3.1)	
Promis Sleep Disturbance[a]		
mean (SD), range	29.06 (7.81), 10-40	
T-score mean (SD), range	61.66 (9.03), 38.1-77.5	
UK reference T-score (Klapproth et al., 2022)	51.26 (8.97)	
Appraisal of caregiving (FACQ-PC)	Mean ± SD	Comparison parent data (Collins, 2020) Mean ± SD
Caregiver strain	3.9 ± 1.1	3.9 ± 0.7
Positive appraisals	3.7 ± 1.1	4.2 ± 0.7
Caregiver distress	3.5 ± 0.9	3.3 ± 1.0
Family well-being	4.0 ± 0.9	3.6 ± 0.7

[a] 50 is the population average; lower values indicate better function.

In the appraisal of caregiving scale, caregivers reported high levels of caregiver strain (index of burden) and distress as well as family well-being and positive appraisals. The mean scores for each subscale were calculated (range 1-5) with higher scores indicating higher levels of the construct being measured. These results are compared with those presented by Collins (2020), which related to a sample of parent caregivers (n=16 fathers and n=123 mothers).

Health related quality of life data is presented below. The number of participants and proportions of categorical responses for the five dimensions of the EQ-5D-5L (mobility, self-care, usual activities, pain/discomfort and anxiety/depression) are presented. Each participant provided a 1-digit number between 1-5 for each dimension (no problems=1 to extreme problems=5). The digits for each dimension were combined for each participant to describe their 5-digit health state (where 11111= full health and 55555= worse than death). Health states were then converted into a single index value using a UK specific value set (EuroQol). The mean index score for the sample was 0.89 (1= best possible QoL). Participants were also asked to rate their health on the EQ-VAS scale. The mean EQ-VAS score was 71.91 (0= the worst health you can imagine and 100= the best health you can imagine). Both scores were similar to UK reference scores which can be seen in the table below.

Table 9; EQ-5D-5L results including frequency and proportions for each dimension and level, VAS scale and index score

Dimension	**Level of problem**				
	Level 1 No problems n (%)	Level 2 Slight problems n (%)	Level 3 Moderate problems n (%)	Level 4 Severe problems n (%)	Level 5 Extreme problems n (%)
Mobility	30 (93.8)	2 (6.2)	0	0	0
Self-care	32 (100)	0	0	0	0
Usual activities	31 (96.9)	1 (3.1)	0	0	0
Pain/ discomfort	24 (75.0)	6 (18.8)	2 (6.2)	0	0

Anxiety/ depression	8 (25.0)	8 (25.0)	14 (43.8)	1 (3.1)	1 (3.1)
VAS score (mean, sd)	71.91 (13.45)				
Index value mean (95% CI) [range]	0.89 (0.86-0.92) [0.648-1.000]				
UK reference index value mean (95% CI) [range]	0.82 (0.80-0.83) [-0.573-1.000]				
UK reference VAS score mean (SD)	71.63 (21.21)				

5.1.1 Summary of quantitative findings

Thirty-two fathers, aged between 36-54 years, took part in the survey. The majority of fathers were White and were born in the UK. Half of the fathers described themselves as their child's primary caregiver, with the remaining half either sharing responsibilities with their partner (n=9) or describing their partner as their child's primary caregiver (n=7). Twelve fathers described their employment status as home/caring duties, with the remaining fathers being in full-time (n=14) or part-time/casual (n=5) employment. The children, who were aged between 3 and 23 years had a range of diagnoses. Most children required significantly more care most or all of the time, compared with children of a similar age. Most of these children were hospice users (n=28).

Fathers' appraisals of caregiving suggested high levels of distress and strain, as well as high levels of family wellbeing and positive appraisals. However, positive appraisal scores were lower, and family wellbeing scores higher, than those reported by Collins (2020) as well as those reported in adult palliative care (Cooper et al., 2006).

On average, participants reported similar health related quality-of-life compared with UK normative data (Klapproth et al., 2022). For mobility, self-care, usual activity, domains, few fathers reported any problems. There were some slight problems reported in relation to pain/discomfort. Mental health problems were more prominent, with 16 fathers reporting moderate, severe or extreme anxiety/depression. Fathers' PROMIS sleep T-scores (61.66) were higher (indicating worse function) than the UK reference score (51.26) suggestive of issues with sleep quality, falling asleep, restless sleep, and satisfaction with sleep.

5.2 Qualitative study findings

12 fathers of 15 children with a life-limiting condition, completed interviews, 9 of which were initially recruited to the survey via children's hospices, 2 via an NHS children's hospital and 1 via social media. Three further fathers provided their contact details for me to contact them about an interview but did not respond further. All interviews were carried out remotely via Zoom.

The fathers that took part in the interviews were aged between 39 and 51 years and resided across various regions on the UK. This included fathers of children who did and did not have support from a children's hospice. Most fathers were in full-time (n=7) or part-time employment (n=1), and the rest (n=4) were full-time caregivers. The majority of the sample were White, and one participant was from a minority ethnic group. Their children were aged between 3-23 years, 7 were male and 8 were female. The children had a range of diagnoses including congenital (n=2), genetic (n=5), neurological (n=6), metabolic (n=1), and cardiac conditions (n=1).

Thematic analysis resulted in 3 main analytical themes; (1) precarity; (2) feeling understood; (3) trajectory of child's illness and the importance of temporality. These themes are described in detail, with exemplar quotes, below. These themes describe fathers' experiences, which are so very often centred upon an overwhelming sense of uncertainty. Their own health and wellbeing is not a priority, though is affected by their child's illness and associated stressors. The health and wellbeing, and support needs fluctuate, often in parallel to their child's deterioration or improvement. This leads us to the final theme through which the importance of trajectory and temporality in fathers' experiences is highlighted.

5.2.1 Theme 1: Precarity

Precarity describes the uncertain, unstable, and inflexible nature of fathers' day-to-day lives. There was a real fragility to their accounts of their daily routines, that relied upon things 'going right'. The theme is particularly focused upon the impact of, and challenges associated with, maintaining uncompromising care routines for their child, which allowed for little variation or spontaneity in fathers' everyday lives. The precision with which fathers described caregiving routines really demonstrated the inflexibility of these routines. These routines were central to fathers' accounts; they were non-negotiable responsibilities described as part of a closed and prioritised system, with other things, like social activities, work, family time and hobbies, on the periphery.

The nature of precarity was multi-dimensional and reciprocal, between this caregiving system, i.e. the inherently precarious nature of life-limiting conditions and care provision, and those peripheral factors i.e. fathers' 'normal' life events and activities. Fathers experienced precarity not only in relation to the practicalities and logistics of being caregivers, but in their own emotional and psychological states. There was a real sense that fathers were 'at capacity' both practically and emotionally, with little scope to deal with additional stressors. This theme covers the various individual origins and impacts of precarity, and the subsequent all-encompassing sense of precarity experienced by fathers.

In the interviews, we often started with what fathers' day-to-day lives look liked, which is where this sense of precarity started to unfold. They described extensive daily medication and feeding routines, that had to be administered at certain times of day, and overnight, requiring a lot of practical and time-consuming work and high levels of organisation. The inflexibility of these routines was particularly prominent.

"Her food runs over 13 hours, so to get everything done, she has to be fully switched on by 6pm. She has to have all her drugs run through, so starting about quarter-past-five so they've had a good 40 minutes or so. So, she's fully plugged in which means she has to sit until she falls asleep, so she has to sit some nights for 2 hours and she gets bored and frustrated, so we have to sit with her and keep her attention. That finishes by 7 o'clock the next morning" (father 1).

Not only did fathers highlight the time-consuming nature of such routines, but they also described the complex nature of such e.g. the importance of sterile care.

"I go into her room in the morning for a quick handover and let the night staff go and then we have a full-on sterile nursing care. She's on 5 different medications, which takes an awful lot of preparation and quite a long time to give through infusions" (father 10).

One father described the amount of medications his son needed, alongside emphasising that he and his wife did it all themselves to avoid the risks of mistakes.

"He also has chest physio every morning and he's on a ventilator overnight. We take over from the nurse at 8am and do his personal care like washing, pad changes, getting him dressed. It's a slow process. His medication preparation is a significant task. We don't delegate that to anyone, simply because it gets so complicated to oversee it and keep track of all his meds. He is on a long list of medication, about 10 or twelve different types. At night it's all of that in reverse. I do daily stock checks of his meds and often have to chase up pharmacists for stuff we're running low on" (father 5).

Daily routines were centered around meeting these caregiving responsibilities, which fathers often had to meet alongside other activities such as taking their child/ren to school and their own work commitments.

"If I have to be in the office at least three times a week, which means that if it's a day I'm in the office it makes it a little more difficult because my partner is by herself, and we have to get [sibling] to school" (father 2).

Similarly, one father described how difficult it could be to get day-to-day tasks done given how busy he was.

"Then you've got all the other stuff, regular day to day stuff. There's just always something to do" (father 4).

There was a real fragility and threat of collapse to fathers' accounts of caregiving routines, mainly due to the inherent instability of the child's condition itself. There was an awareness from father's that despite applying high levels of control to their daily lives, that was essential to meeting the caregiving needs of their child, everything could change if their child became unwell. Fathers remained hypervigilant for any change, with uncertainty becoming normality.

"Last year she was in hospital seven times from September to March, and basically she'd get a cold or a little bug, but when she gets a cold she'll throw up and because then when she throws up she gets dehydrated she has to go to hospital to get rehydrated. So almost on a three week... she'd be well for a week, then go to hospital for a week, and that was kind of all last winter. Last winter was probably one of the worst, apart from when she was born" (father 4).

This hypervigilance was essential when doing things that fathers knew had affected their child's condition in the past.

"Getting her out of the house is really tricky and really hard work. You've got to pre-heat the car to make sure it's the same temperature as the room she was in. You have to run her out of the house like a rugby ball, through the outside and into the car. You have to hope that whatever room she's going into after the car is about the same temperature. Any failure in this will cause a seizure" (father 10).

Slight changes in the child's condition caused anxiety for fathers as this was often a sign that things would deteriorate further, sometimes requiring a hospital stay. Deciding if their child was unwell enough to go out could be challenging but fathers did often try to mitigate the risk of further illness by cancelling plans such as school or playgroups.

"So in principle, they have a Monday play morning where you can take your child and a carer plays with them, and you can have coffee; we absolutely want to take her but it doesn't happen as often as we'd like. She only attends about a quarter of the time due to being too unwell" (father 10).

For working fathers especially, there was a real rigidity of routine and an overwhelming sense of constant responsibility whether that be caregiving or to their employer.

"So, I get into the office for 9am, I leave at 7.20am. The bus comes to pick [child] up at 8.30am and [sibling] gets the bus to school too. So anyway, up until 9am it's quite eventful, even more so if she's had seizures in the night because then we're all tired. The school bus brings [child] home for 4.30pm and I look after her while my wife prepares tea. [Child] is put on a feed for teatime and we have meals together. At around 7pm we put [child] to bed. It takes about an hour" (father 2).

They would often resume their caregiving responsibilities straight from work, to give their partners a break; a 'shift' like approach, particularly seen with overnight caregiving responsibilities.

"And I get home, obviously after a long drive and a day in the office, and then I'm kind of looking after [child] from that point onwards" (father 2).

Some of this stress was eased by changes to working patterns during the COVID-19 pandemic, which is touched upon a little further below. Regardless of the extent to which fathers were directly involved in caregiving or night-time routines and monitoring, the precision in timing with which these activities were portrayed, was common across fathers' accounts.

"What's hard though, is that equally some people have quite demanding jobs, they get home and it's like ah, right, now I can relax. I usually can't most evenings I have to stay up to hand over care to [child's] care. I have quite a few drugs to prepare and feeds and things" (father 11).

Establishing routines often relied upon daytime or overnight carers for their child. Much of the insecurity in fathers' accounts stemmed from a lack of, or withdrawal of this type of support.*"We used to survive on three or four hours of sleep per night", which was just awful. Now, we get better sleep but that's all changing again because his carer that he's had for three years has just quit and left"* (father 6).

This was particularly the case during the COVID-19 pandemic and was evident through accounts of this time, during which fathers felt abandoned, lonely, and overwhelmed.

"During the pandemic, it was tremendously gruelling to try and look after her under those conditions, with so little support. My wife is a very law-abiding person so we really kept to what we should be doing and that was, yes, tremendously hard. We asked the GP if they could help, so they sent a social prescriber round who said, "there's absolutely nothing I can do for families like yours. There used to be before the pandemic, but it's all dried up now"" (father 10).

Another father highlighted that the majority of his child's carers were unable to help during the pandemic. This father uses the term '**dropped** away' which highlights the sudden abandonment felt by many of the fathers in this study.

"We locked down. Pretty much all the carers dropped away and there was just me and my wife. It was isolating" (father 6).

The pandemic also meant that many fathers had started working from home or had started to work more flexibly. For some, this seemed to mitigate some of the tensions associated with inflexible and overwhelming care routines, allowing fathers to support caregiving throughout the day, reduce their commuting hours and allowed a little more 'give and take' when things did not go to plan. Fathers also recognised that had it not been for the pandemic this flexibility may not have existed.

"The pandemic has increased flexibility and it is very helpful. I think a lot of parents find that.

I think for us it's particularly helpful. The fact that I'm at home working rather than in the office, not that my commute is massively long or anything, but the fact I'm at home in a way that I wasn't means that I can help out during the day so that my wife isn't stuck with it all" (father 11).

Another father described feeling happy that he was able to care for his child whilst also attending work meetings. However, he also highlighted that there were tensions associated with trying to manage his child's needs during working hours.

"I feel quite happy in that I don't feel like there should be any stigma with having your child in an online meeting. It's not always easy when [child] is in pain and it's not easy for the people I'm on the call with. The job is being done but with a great deal of strain. I anticipate that things would have been very different had it not been for the pandemic" (father 10).

However, for some fathers working from home had been particularly challenging, highlighting the constancy of their caregiving roles and leaving even less time to focus on other things, adding to the overwhelming nature of their daily responsibilities.

"Yes nine to five work. When I started working remotely, there is no actual nine to five work because it's just like "yes you're at home to you can look after this and this and this". There's so much to do" (father 12).

The maintenance of such routines, alongside the threat of collapse, was both physically and mentally exhausting for fathers.

"We are physically and mentally exhausted. We had no resources left, I had nothing" (father 1).

They described having little time to relax due to both the practical demands of caregiving and the inability to 'switch off mentally'. This lack of time to relax included a lack of time to take part in regular family and household activities, spending time with partners and other children and having time to do things for themselves.

"This stuff just sucks time away, which means your life is just fire-fighting and it's very hard to get on top of stuff. Me and my wife don't get to sit down before about half past nine and we watch tv for thirty minutes and then go to bed. That's our day, thirteen or fourteen hours of drudgery and then thirty minutes of free time, but we're knackered" (father 7).

This sense of overwhelm also relates to challenges they experienced in coping with other stressful life events alongside their child's illness, and the potential that these events had to impact upon fathers' ability to care for their child. One father described his concerns about his parents needing extra help in their old age, but worried that he wouldn't be able to provide the care they needed because of the care he was already providing for his child.
"My mum and dad are getting on a bit too... I just can't be there as much as I need to be. There isn't enough time and I just don't have it in me... which worries me as there's no one else" (father 6).

Fathers also described fears about job security and finances. Fathers described these events as normal life events, that on their own would indeed be stressful, but on top of their caregiving responsibilities were exceptionally difficult to manage both emotionally and practically.

"I'm well aware that it's on me. I do okay money wise but if I lost my job tomorrow things would quickly become very difficult. It's a lot to manage stress wise... knowing that the security of my family, the house, everything, is down to me" (father 6).

One father who had a good relationship with his boss, and a flexible job, was particularly anxious about changes his employer was making:

"They're making some changes at work I think my job is safe but I don't know. There are other jobs coming up in the company but I'd have to apply and it would mean relocating which we obviously can't do easily. I need the flexibility that I have at the moment. and the thought of having to start a new job with everything that's going on at home. it's a very anxious time" (father 8).

This theme highlights the precarious nature of caregiving; an over-arching sense of uncertainty, coupled with the inflexibility of routine which could be overwhelming for fathers, making additional stressors hard to manage. Fathers' lives seemed to be particularly vulnerable to these additional stressors.

5.2.2 Theme 2: Feeling understood

This theme explores the health and wellbeing of fathers and the barriers they experienced in addressing their own support needs. Fathers described a range of concerns, mainly in relation to their mental health and wellbeing, though could find it difficult to express these to others. This was not only due to the lack of opportunity to do so, but apprehension surrounding how their concerns would be interpreted, meaning they were often left unaddressed until they became urgent. Conversely, through appropriate and suitably timed care and support, fathers felt understood, not only in relation to their own wellbeing but in their role as caregivers.

There was some variation to the way in which fathers discussed and managed their mental health. Some had received specific diagnoses, such as anxiety and/or depression, in response to their child's condition. Some had not had any formal mental health diagnosis, but described low mood, stress, anxiety, and mental exhaustion, the impact of a lack of sleep.

"In terms of mental health, I struggle from low mood. It's definitely low mood as opposed to depression" (father 10).

Fathers did not often discuss their physical health but when they did, they mainly discussed physical exhaustion and back pain related to the physical exertion of caregiving including lifting and carrying their child and their equipment (or often lifting their child because of a lack of equipment.

"I'm enduring it. Exhaustion is at the forefront of it. Because trying to monitor [child] 24 hours a day is very very hard and has meant that I have stopped exercising. I'm getting ill more often, backaches, headaches" (father 10).

Many fathers described being able to seek support for their psychological health if they needed to. For some this was through a good relationship with their GP.

"I guess that if I felt as though I was going into a mental health crisis then I would talk to my GP. If that's the type of thing I needed, I could progress that but it doesn't seem like a pressing concern at the minute" (father 10).

Others did not define a pre-established route to psychological support but knew that they would seek help should they need to.

"I'm fully aware that if I'm struggling, I need to speak to someone and I know that I can do that" (father 1).

One father highlighted specific means of support through which he knew he could talk to someone fairly quickly.

"If I needed it, I'd go back to the wellbeing service or the children's hospice. They would be my first port of call. It's the quickest route to being able to talk to someone" (father 8).

With respect to mental health, fathers found confidence in being able to ask for very specific means of support, for example a prescription for anti-depressants from their GP, rooted in an understanding of what worked well for them and when. One father described 'keeping an eye on his mental health', deciding whether he was feeling understandably stressed or whether his depressive symptoms needed to be addressed through his GP.

"I really try to distinguish between distress and depression where the circumstances I'm in are naturally going to be producing distress and to not feel like that's the wrong thing but to try to watch out for feeling more low than the situation warrants" (father 10).

This ad-hoc type approach to support seeking was complimented by their ‘getting on with it’ approach to caregiving, finding individual coping mechanisms, and fathers’ lack of desire to ‘dwell on things’, and for some was helpful. Conversely, it meant that fathers could be faced with unclear thresholds for support, ‘putting up’ with things and leaving concerns unaddressed until they became urgent. One father described his experience of getting to 'breaking point'.

"First, I looked at people with mental health problems as weakness, and it's not at all is it, it's actually the opposite. If you're the sort of person that doesn't push yourself through then you never actually get to breaking point. I think I pushed on and pushed on and pushed on to the point where I just couldn't keep going, it was like a fuse had blown" (father 5).

As well as the general lack of opportunity for men to discuss their feelings and worries, these tensions were sometimes rooted in fathers’ references to gender norms and needing to be strong for others.

"Us dads don't talk. I know an awful lot of dads out there that don't because you always feel that you're expected to be strong. As a dad, it's your job to protect your child and your entire family. I think a lot of dads struggle with that. We just want to make sure that people think we're strong for everybody else. You can only do that for a certain period of time before you cave in" (father 8).

For those who did not have specific mental health concerns or diagnoses, but suspected that additional support would be beneficial, there was less support available. Fathers found generic interventions like short-course psychological therapies, anti-anxiety, or anti-depressant medications to be inadequate in addressing their needs as caregivers. One father highlighted the risks of such interventions:

"I found it quite painful honestly and that the six to eight sessions available, that it wasn't really safe in the scope of such a short period to go into the depths of what was going on and also in the time after the sessions. I would be less functionally available to the family because of what I had to go into and come out of" (father 10).

As well as safety concerns, fathers described support that just was not able to address their needs as caregivers. One father described not having any help that was applicable outside of the therapeutic setting:

"The best she could come up with was some deep breathing exercises. The following week she was telling me about therapeutic doodling, just get a pen and paper and doodle for a bit. I was like I'm not entirely sure that's going to answer my questions. I've told you everything but I haven't got any tools to go away with" (father 1).

Fathers highlighted the generic nature of such interventions.

"The problem is that she sort of discounted it, it's like, "oh yeah, well, you're really busy aren't you?" Its like "well yeah, but I probably have a burden far greater than most people you're dealing with". So it's a bit of an generic tool that's not addressing the problems of [child's] condition (father 7).

Feeling understood, and being recognised, was important for fathers and there is a distinction to be made between being misunderstood versus ignored. The inappropriate support offered to fathers led to them feeling misunderstood. Related to this was one fathers experience of having the impact of his mental health being misunderstood with a social worker suggesting family coping, and their child's safety, could be improved by taking antidepressants.

"There was about four or five months where there were quite difficult conversations with the social worker it was felt that our distress was a danger to [our child] because it meant that we were less likely to be coping as a family and therefore needed to be fixed with antidepressants, as if that was going to fix it" (father 11).

Professionals involved in their child's care were described as 'assuming that mothers were primary caregivers' and tailoring support accordingly. Fathers felt that their dialogue with these professionals was limited and occasionally challenging, partly due to these pre-existing gender biases, partly due to the nature of their interactions with these professionals, i.e. at their child's medical appointments during which there was a lack of time to discuss anything other than their child.

"He had a really strong epileptic seizure and they just said he needed a mummy cuddle and I said or a daddy cuddle because everything, literally everything is female centric. Is that the right word? And quite a few times I've said I am almost forgotten. That sounds like I'm pleading poverty or whatever it is but it feels like I'm just a spare part in the background. But me and [partner] are 50/50 on this whole thing. She couldn't do without me and I couldn't do this whole thing without her. So we're definitely a 50/50 split but definitely more people ask about [child]. Only a couple of people medically, probably one person would only ask me how I'm doing in six years, a medical person" (father 6).

.

Another father reiterated a mother-centric healthcare system.

"Our life is up to their condition to be honest. Everything is related to mums but I know my struggles. Nobody cares about fathers" (father 12).

There was also a lack of desire from fathers to address their concerns, particularly with their partners, due to a fear of burdening them with additional worries. Fathers wanted to appear to be strong and to be coping well, and described feeling guilty if they were struggling, describing their struggles as unwarranted, particularly if they were not primary caregivers.

"I don't know why. Maybe I personally don't offload because I feel like I don't want to burden other people, or it feels like I should be able to cope better" (father 6).

This was also related to gender based role function expectations.

"Maybe that role is still there, the bloke is out at work and the woman looks after the house and child kind of thing. So the focus is on the mum, quite rightly, and it's your job to protect the family" (father 8).

Fathers found it difficult to talk to friends about their experiences, partly due to not wanting to burden them.

"I get frustrated but sometimes I'd rather not talk about it because I really don't want to

burden other people" (father 6).

Fathers mainly felt that friends did not understand, and it could be emotionally taxing to keep talking about their child's illness, especially with those who could not understand to true extent of caregiving.

"Initially, I would try to keep everyone updated with individual phone calls but it was really counterproductive. I'd end up repeating the same grim old facts to everyone and I'd feel sad and they'd feel helpless" (father 10).

Furthermore, some fathers did not want to talk about the challenges associated with caregiving and their child's illness, because friends would not know what to say and could make unhelpful comments. *"That's not anything badly on my friendship with my best mates, I just don't feel that they would fully see where I'm coming from or they would try and say things just to help"* (father 8).

This was particularly difficult for fathers when friends or family would try and offer a solution or hope to the situation.

"Sometimes you've spoken to family members, they'd say things like "Oh I know somebody who had epilepsy and they stopped having it when they were 11", which to be fair is an ordinary persons experience of it. I think people just don't see it, always looking for solutions or to reassure me that things are going to be okay. But you know it ain't going to happen" (father 2).

This theme highlights the multitude of barriers that fathers faced in addressing their concerns, particularly around mental health and wellbeing. Waiting until they needed urgent mental health support, meant that fathers were less able to explore long-term and sustainable approaches to looking after their health, as getting well enough to care for their child was the priority. Fathers could find it difficult to discuss their concerns with friends and family, who often did not recognise or believe the difficulties of fathers' experiences.

Fathers described the close relationship between their child's health and their own mental wellbeing. The inextricable links between the two meant that fathers would often struggle the most when their child's health was in crisis. Again, in these instances there was little time for fathers to explore their own needs as they were so focused upon those of their child and family. These quotes lead into the third and final theme focused on trajectory and temporality.

> *"I can certainly appreciate that there may be benefits [to therapy], but I wonder whether things are just too acute at the moment for it to be a good space for reflection and trying to make sense of things. I think we're too close to crisis for it to be feeling steady enough to explore feelings"* (father 10).

> *"I feel like I need time to heal, it's like a healing process. But the situation we're in and the demands that we have with [child] makes that impossible. I want there to be a time when I'm off medication but it feels like I'm always going to be on it"* (father 5).

5.2.3 Theme 3: Trajectory of child's illness; the importance of temporality

The final theme surrounds fathers' experiences in relation to the trajectory, or course, of their child's illness and the importance of the temporality in their accounts. Fathers described past traumas that they felt unable to resolve, as well as fears related to the future of their child's health, and life, contributing to a real sense of existential uncertainty. Of prominence was the way in which their past experiences, and future expectations, shaped the way in which they functioned in the present and the way in which fathers repeatedly restructured their wider temporal expectations, throughout the course of their child's illness. This theme is split into 4 sub-themes; (1) "Realising there was something wrong" describes the point at which fathers knew that their child was seriously ill. This was sometimes at the point of a formal diagnosis, but often, fathers had known their lives were going to change before that. It relates to those very early days of their child's illness, fathers' accounts of their journey to diagnosis and some of the immediate impacts of such; (2) "lost futures and re-shaping expectations" explores the way in which fathers talked about loss of the futures they had envisioned for themselves. They were required to let go of things that they had once felt were important, and re-shape their multiple roles in the context of their child's illness; (3) trauma on trauma describes the cumulative effects of distress on the wellbeing of fathers. Having little time to address traumatic experiences meant that fathers carried these with them, making it challenging to cope with further distress; (4) "anticipating suffering" describes the fear fathers expressed in relation to their child's future health, and death.

5.2.1.1 *Realising there was something wrong*

Fathers described their experiences of receiving their child's diagnosis. For some it had been many years since diagnosis, for others a shorter period had passed. Regardless of these differences, there was a real clarity in fathers' accounts. They described the traumatic experience of diagnosis, emphasised by this vividness with which they recounted it. For many, particularly those with rare neurological disorders, families had waited for some time for a diagnosis.

"We took her to A&E and they thought it was some kind of wind, some kind of gastronomic issue. So she was put on a ward for those issues. They monitored her there and they were beginning to get concerns that it wasn't wind. They did a brain scan and I wasn't there because I was working and I thought "oh it's just a general procedure to rule things out". Anyway, the next day the report came back and the consultant... [sobbing] sorry. From there we knew there was something seriously wrong neurologically" (father 2).

Fathers described this period as one of uncertainty and ongoing attempts at self- reassurance that nothing was wrong, readjusting these hopes at various stages, based on new information.

"So I think it's been a bit of a journey in terms of reconciling ourselves- well, me in particular, about- maybe thinking to myself in the beginning "Oh it will be okay, it's just this" to "Well, okay it's just a moderate developmental issue that she'll grow out of" until the realisation that actually it isn't going away, it's there and she's going to have to live with it and we're going to have to live with it" (father 10).

For children that had been diagnosed during the Covid-19 pandemic, fathers described feeling abandoned by professionals. The shock and uncertainty associated with diagnosis was particularly intense for these fathers. There was little to no immediate practical or emotional support following on from diagnosis, and fathers felt a complete lack of compassion from healthcare professionals. They described a brutality in their experiences; feeling like they were given the 'worst news of their lives' and then 'just left to get on with it with no clue of how to do that'. Not only did this feel extremely isolating but seemed to set a precedent for how fathers came to expect support from professionals throughout the course of their child's illness.

"This was at the very start of the pandemic. We were given the news and then kind of ushered out of the door of [children's hospital] as they were evacuating.

We were left to Google to find out about the condition that way" (father 10).

As well as a complete lack of information, fathers also described a lack of compassion in the delivery of information.

"We were copied into a medical letter. They basically sent us a letter saying "there is something seriously wrong with your child". There was no compassion in it at all. We were so upset that it was broken to us in this brutal way" (father 7).

Not only did they often have to fight for information but this was often followed by a lack of support.

"From here.. if I were to characterise the last ten years, it's been the same... a never-ending war to get support" (father 7).

5.2.1.2 Lost futures; reshaping expectations

Fathers experienced a deep sense of loss related to opportunities for their child, as well as to the life that they had envisioned for themselves as fathers, the type of parent they thought they would be and the family life that they thought they would have. Fathers described uncertainty in relation to their new roles, unsure of how to reshape their perceptions and expectations of what it meant to be a good father, employee, partner, and friend in the context of their child's diagnosis. They described that a certain level of acceptance was required.

"I think, once you begin to reconcile things... we'll just enjoy [child] for what she is. At first when we went out in the wheelchair, we were a bit self-conscious about people looking at us. You thought, oh god, they're looking at us and thinking "oooh, he's got a disabled child" and they feel sorry for you. Somehow you feel as though you have let someone down. But anyway after about 6 months I realised no-one was looking at me" (father 2).

This sense of loss at diagnosis was compounded by the ongoing and frequent losses that fathers experienced at various timepoints often in relation to seeing the pace of development in other children.

"We used to take her to baby groups and it was just so disheartening to see other children developing and she just obviously wasn't" (father 2).

Similarly this included seeing their friends' children as well as activities that their friends were able to do.

"You just recognise that. friends who have had children around a similar time to me. They have been able to reclaim a bit of the life they had before children at a much quicker pace than I have been able to do or am, frankly ever likely to do. It isn't ideal but it is what it is" (father 11).

Losses were sometimes only realised when milestones, family occasions and activities were not reached and was often in comparison to what healthy siblings had been able to do.

"Obviously, seeing that they won't develop like [sibling] did is really hard" (father 9).

Reshaping or replacing these expectations, to allow for the presence of this uncertainty, was fundamental in fathers coming to terms with their child's diagnosis and what this meant in the broader context of their lives. The uncertainty associated with living with long-term palliative care was difficult for fathers, who had to balance normality with a desire to give their child the best life possible.

"If your child has 6 months to live, you spoil them rotten. You go to Disneyland, you do the Make a Wish, you meet Ed Sheeran, whatever. You make it count because it's 6 months. But when it's long term, you can't spoil them every day, you have to almost put it to the back of your mind and carry on like it's not there" (father 1).

Learning to live with uncertainty was an iterative and ongoing process throughout the course of their child's illness and fathers experienced flux in their acceptance and ability to cope. This flux was evident in fathers' accounts of their own mental health (theme 2) as well as being strongly connected to the sense of precarity in theme 1. They described periods of real satisfaction with life, during which their child was relatively well, and their family was able to function regularly. This satisfaction was inextricably linked to the current state of their child's health, but other factors impacted substantially upon fathers' ability to cope, most notably their experiences of trauma and unresolved trauma.

5.2.1.3 "Trauma on trauma"

Fathers described the cumulative impact of 'repeated traumas', 'trauma on trauma' and, in relation to distressing situations, 'just one thing after another after another'. Fathers felt that many of their experiences remained unresolved, having little time or knowledge of how to process them, meaning that subsequent challenges were even harder to navigate emotionally and psychologically. For many this began at diagnosis, as described above. For many the trauma came from the diagnosis itself, for others it was the fear and abandonment they experienced following on from their child's diagnosis.

"I think the key thing is that in that 5 months in hospital we never had the chance to process what had happened because you just try to get from one day to the next. She was in intensive care, she had two respiratory arrests, she had a cardiac arrest, she had three surgeries. You just get through from one day to another and then all of a sudden you're back living in the real world and it's quite scary" (father 8).

Another father described thinking that therapy would be useful, in order for hi, to go over the trauma experienced leading up to child’s diagnosis. There was a sense in many accounts that going over past experiences could be useful but would just not be feasible now given concerns related to how it would affect them or purely related to time-constraints.

"I think I just need to go back to all the stuff that happened when she was born. Its all just so intense and traumatic, just the whole thing. I would love to be able to go back and have full-on therapy and go through all of that stuff when she was younger. But because of all the stuff happening now you just don't have time or space to do that" (father 4).

Fathers described the speed at which caregiving responsibilities took over their lives. The practicalities of caregiving, coupled with busy appointment schedules, house adaptations, meeting new care teams, and chasing up services, which were extremely stressful in themselves but also meant that there was little time for them to process the trauma they described as having occurred previously. Living in the moment day-to-day was the only way in which they could meet the needs of their child and learn how to provide care in a way that worked for them.

"It takes years to get your head round what you're doing, what you've got and that does keep changing. You're always focused on the next step, but then you look back and go "God, that was blumin' awful. We had a really bad time didn't we?" (father 7).

Fathers described the trauma of seeing their child regress developmentally, which was in conflict with fathers' expectations of the future.

"When he went to school, he was walking then. But the thing is with his condition, you get a massive decline in muscle strength, so then he was in a wheelchair. You're dealing with that" (father 7).

Fathers highlighted the emotional impact of seeing changes in their child.

"She used to enjoy being read to, now she doesn't. She lost recognising her cuddly toy that she goes to bed with, she's lost recognising her name. She used to be able to recognise my wife and I when we came into the room, but she doesn't anymore. I mean she knows who we are, but she doesn't greet us in the same way. One thing that I found particularly sad, is that she has stopped smiling. It has been a struggle" (father 10).

5.2.1.4 Anticipating suffering

The final sub-theme is very much intertwined with the concept of trauma-on-trauma, as it deals with the anticipated distress that fathers described in relation to their child's future and decline in condition, including their death. The hypervigilance that fathers described in relation to their child's symptoms resulted from an awareness that their child's condition could deteriorate at any moment.

"[Child] is getting older and the condition itself has the potential to be life-limiting because she's got a central line which we've already had replaced several times, so every time she goes in for a line replacement it becomes this "will she come back from this one? Is she coming back with a new one or are they going to come out to the waiting room and say, we can't do it"? That's where life gets painfully difficult" (father 1).

Fathers highlighted the constant presence of their child's mortality, alongside inherent uncertainty and fear, particularly when their child needed to be admitted to hospital.

"It's constant, absolutely constant and the thing which I always find, this is when I get quite tearful, that with all this stuff you're constantly thinking about when [child] is going to die. When she could die. That's always there in the background. She's completely healthy now but you're always thinking she could die. This could be the moment when she doesn't come out of this, which could be a really long and painful process in itself. She won't just drop down dead. It will be long and drawn out" (father 4).

Fathers also explicitly acknowledged that they knew their child was going to die and support directly related to bereavement could be useful.

"We know there's trauma coming at us and there's nothing much we can do about it, we just know it's coming. And I think this is one of the things we think there should be is pre-bereavement classes" (father 1).

For fathers of children with a progressive disease, there was often a clearer sense of when their child might die, but this sense of uncertainty was still present.

"Something is going to happen very soon. The average life expectancy is 15 and he is 15, so we're completely in the unknown, there's no end in sight as such, but he's declining all the time" (father 4).

For some fathers, the concept of their child's death was too hard to try to make sense of and they wanted to know as little about the future of their child's condition as possible.

"I suppose we'll cross that bridge when we come to it... of course you think about it but you just kind of have suppress it otherwise you lose sight of the here and now".

This conflicted with fathers' desire to reduce uncertainty and be prepared and informed about their child's condition. This was particularly prominent in fathers' accounts of seeking support from those with shared experiences. The emotional and practical support that fathers received was useful, though there often came a point when fathers withdrew from this means of support, as hearing what was most likely going to happen to their child was too difficult.

"I've tried that, it's pointless; we're very aware of the problem and we're very aware that there isn't a happy ending. So, great what now?" (father 7).

There was nuance in the way that fathers discussed their own fears about the future, what their own lives and roles would look like, how it would affect their own wellbeing and that of their family, compared to the suffering they anticipated and feared for their child. This again highlights the relationship between child and family health and wellbeing. It had also forced fathers to consider their own mortality, realizing that their child may outlive them. This caused major concerns about who would look after their child in the future. It also relates to this constant adjustment of expectation in fathers' accounts.

"You don't like thinking these things, but, like I said at the beginning, you get to a certain age and, I'm over 50 years old now, so I'm aware that things do start happening and you've got to be on the lookout for them. That worries me, I'll be honest" (father 2).

5.3 Integration display

Below is the integration display that shows key qualitative and quantitative findings side by side. As this was a qualitatively focused mixed-methods study, the findings are organised around the qualitative themes. Relevant data is presented in the appropriate columns and allowing further inferences to be made, which are then also explored in the discussion.

Table 10; Integration of qualitative and quantitative findings

Overarching analytical theme	Qualitative findings (examples)	Quantitative findings	Narrative summary
Precarity in fathers' everyday lives	*"I work mostly looking after her at night-time, I'm looking after her at morning time. After we send the kids to school, I have to work"* (father 12). *" It's just the level of appointments and the... on medical and educational side plus also the unpredictability of it all"* (father 11). *"I'm well aware that it's on me. I do okay money wise but if I lost my job tomorrow things would quickly become very difficult. It's a lot to manage stress wise... knowing that the security of my family, the house, everything, is down to me"* (father 6). *"There was a very difficult incident about six or seven years ago when [child] ended up in intensive care as a result of a series of complications. I had to disappear, I couldn't work"* (father 11).	Stopped work to care for child (n=11), Reduced hours (n=9), changes to their career path (n=6). Child's primary caregiver (n=16), shared caregiving responsibilities with their partner (n=9). Full-time employment (n=15), Home/caring responsibilities (n=12), part-time work (n=5). Sleep disturbance scores were (higher) i.e., worse than those of the general population (61.66 and 51.26 respectively).	The qualitative findings demonstrate the unpredictable, unstable nature of fathers' and families' daily lives. Fathers had substantial caregiving responsibilities for their child, often alongside employment as shown in the qualitative and quantitative findings. Sleep disturbance, and the constancy of fathers' roles, was further highlighted in the interviews. Fathers described sleep disturbance in relation to worry, as well as the need to be awake to monitor their child.

Overarching analytical theme	Qualitative findings (examples)	Quantitative findings	Narrative summary
Feeling understood in the context of health and wellbeing	*"We are physically and mentally exhausted. We had no resources left, I had nothing"* (father 1). *"It's constantly having to push and fight. In the end what it does to your mental health is you get sick of yourself. You feel mentally and physically drained because you feel that all you're hearing is yourself in a negative way because you're constantly arguing and fighting for things and fighting with these people and asking for this"* (father 2). *"Our life is up to their condition to be honest. Everything is related to mums but I know my struggles. Nobody cares about fathers"* (father 12). *"I just think a quarterly call with a trained counsellor would solve so many of the problems that appear two, three, four years down the line, you know?"* (father 7).	EQ-5D-5L index and VAS scores were slightly better than those of the general population. Caregiver strain and distress were high, as was family wellbeing and positive appraisals. Sleep disturbance scores were (higher) i.e., worse than those of the general population (61.66 and 51.26 respectively). Child's primary caregiver (n=16), shared caregiving responsibilities with their partner (n=9).	The findings of the survey highlight the type of health concerns faced by fathers, which were mainly related to mental health (as shown in the EQ-5D domain scores). Despite the overall EQ-5D index score being high, the individual domain scoring highlights issues related to mental health. In combination with the scores of the caregiver appraisal and sleep scales, and the qualitative interviews, these findings demonstrate the importance of recognising the caregiver support needs of fathers and that a better understanding of their overall health is needed. An important part of feeling understood was through the recognition that positive and negative emotions and experiences could coexist- as highlighted by caregiver appraisal scale and interviews.

Overarching analytical theme	Qualitative findings (examples)	Quantitative findings	Narrative summary
Trajectory of child's illness; the importance of temporality	*"So I think it's been a bit of a journey in terms of reconciling ourselves"* (father 10). *"Just as we got the diagnosis, the pandemic hit and so [children's hospital] gave us the diagnosis just as they were heading out the door because they were evacuating the building so we were left to Google and find out about the condition that way. Given that there was only 800 cases in medical history, there wasn't that much available"* (father 10). *"You keep moving to the next terrible stage. I mean, of course what we're thinking of now, and it's three years away, is what do we do when he leaves school because then we don't have a child who is engaging and in a loving environment...he loves it there, and they're really nice and they're fantastic, but when he's 16 he's out the door, so what do we do with him after he's 16 and what is he going to be like when he's 16? So yeah, we're planning for that, but you know that's going to happen but, yeah, you can't stop it, you know?"* (father 7). *"Something is going to happen very soon. The average life expectancy is 15 and he is 15, so we're completely in the unknown, there's no end in sight as such, but he's declining all the time"* (father 4).	Average age of child 11.7 years (range 3-23 years). 11 fathers had stopped work to care for their child, 9 had reduced their hours and 6 fathers had made changes to their career path to care for their child. Most children needed significantly more care, most or all of the time, compared to other children of a similar age.	Many of these fathers had been providing care for their child for many years and many of the children had complex needs (multiple diagnoses) which are associated with unpredictable trajectories. The interviews highlighted the ongoing reshaping of expectations in relation to their child, careers, and wider social lives.

5.4 Summary of key findings; the health and caregiving experiences of fathers

Precarity

The results of the primary study show that fathers experienced various forms of precarity in their everyday lives. This precarity often existed as a direct result of the inherent instability of their child's condition, making fathers' everyday lives unpredictable. Fathers described a real fragility to their everyday routines that relied upon everything 'going right'. They described precarity in their relationships with friends and extended family, mainly as a consequence of feeling misunderstood, feeling like they had to repeatedly explain their child's condition to others, and worrying that their experiences would cause distress to those around them. This unpredictability could be particularly challenging for working fathers who had to navigate role overload. Flexible and understanding employers were important in helping fathers to navigate their schedules and responsibilities.

Feeling understood

Fathers were 'at capacity', both physically and emotionally, making it difficult for them to deal with other life stresses alongside managing their child's condition. This was particularly evident for participants who were dealing with other caregiving responsibilities, e.g. for their own parents, or other stressors such as issues at work. Fathers highlighted the importance of feeling that their health and wellbeing concerns were both understood and taken seriously and wanted their caregiving responsibilities to be acknowledged in the context of these concerns, alongside the recognition that they were individuals with unique support needs. How fathers managed their health was very much rooted in gendered assumptions of health and support seeking.

Trajectory of illness

Finally, the importance of understanding the trajectory of their child's illness when considering the health and support needs of fathers was particularly important. Fathers described fluctuating support needs throughout the course of their child's illness, and how the impact of repeated and unresolved distress, is particularly important to address. The relationship between the health of fathers and that of their child and wider family must also be considered, alongside the routines that they must all adhere to in the face of precarity.

6. The health and caregiving experiences of fathers of children with a life-limiting condition: Findings

In this chapter, I will discuss each thread of the thesis; the survey and interview results from chapters 4 and 5, and how these results build upon the systematic review findings in chapter 2. I will summarise the main findings of the primary study on fathers, with a particular emphasis on the qualitative findings. I will highlight the importance of recognising fathers as parents, and as caregivers, and as individuals. However, fathers can feel marginalised by services and by health professionals meaning that their needs can go unnoticed, perpetuating the view that support is not available to them. This will be discussed in relation to the wider context, building on what we know about caregiving, fatherhood, and parenting, as discussed in chapter 1. I will discuss this in relation to the experiences and health of mothers (chapter 3) and how mothers' and fathers' experiences compare, why this is important in policy and in practice, but equally that it is not so much about comparing their needs, as it is having an understanding that all families operate uniquely. I will consider the implications of these findings for policy, practice, and further research and I will then reflect on the strengths and limitations of the research.

As outlined in chapter 1, the number of children living with a life-limiting condition is increasing (Fraser et al., 2020b) and therefore, so are the number of parent caregivers who must provide care for their child at home. This caregiving is often extensive and can have detrimental effects on the wellbeing of caregivers, making it more challenging for them to provide care for their child. Although mothers are still more likely to be their child's primary caregiver, many caregivers are fathers, who are significantly under-represented in research (Nicholas et al., 2020). This means that what we know about the support needs of parents is mainly based upon the needs of mothers, which is inappropriate given the family centered nature of the paediatric palliative care model (WHO, 2023); to look after their children, all parents must be well, and they must be well supported in their roles. Through this thesis I have also highlighted that although the literature overrepresents the experiences and support needs of mothers, how this has been used in practice is limited. In exploring mothers' experiences of health, and how they want their health to be supported, it has become evident that current mechanisms are insufficient in doing so. Current literature, alongside shifts in parenting roles, demonstrate the need for further research surrounding the potential support needs and experiences of fathers too, but recruitment to research remains low compared to mothers (Nicholas et al., 2020). Therefore, the aim of this thesis was to explore the health, wellbeing, and caregiving experiences of fathers of children with a life-limiting condition which have so far not been explored in this way.

6.1 Conceptual models of caregiving; scope and purpose

In chapter 4, I explored several influential models of family caregiving, underpinned by concepts such as stress, coping, burden, resilience, and burnout (Lawton et al., 1991, Lazarus and Folkman, 1984, Pearlin et al., 1981). We see substantial agreement across existing models, particularly in the centrality of the relationship between stressors, and individual, family, social contexts, and cognitive factors which we know are fundamental to our understanding of the effects of caregiving on health and wellbeing. As also discussed in chapter 4, many aspects of the models remain under-developed, particularly in their application to parental caregivers, and particularly to fathers. Very little is known about the health outcomes of these fathers in relation to caregiving, even more so the contextual factors that contribute to them.

Framing this research around the concept of caregiving, and its impact on health, allowed me to address this imbalance and subsequent gaps in research. The Informal Caregiving Integrative Model (ICIM) defined by the Gérain and Zech (2019) (figure 6) provided an exploratory framework in the design of the study, through which I will also discuss my findings. This model was developed by incorporating important concepts from both informal caregiving (Pearlin et al., 1990, Lawton et al., 1991, Sörensen et al., 2006) and professional burnout (Demerouti et al., 2001). Burnout, as a response to chronic stress, is a tridimensional concept made up of emotional exhaustion, depersonalisation, and personal accomplishment (Lee and Ashforth, 1990), and has so far remained limited in its application to parental caregiving despite empirical evidence suggesting its relevance (Gérain and Zech, 2019). In the model, burnout is a key mediator between determinants and more general outcomes. Appraisal, and relationship with the caregiver, act as key mediators between said determinants and burnout, as demonstrated in figure 6. It is important to reiterate that the model was primarily intended as a tool to assist with the exploratory nature of the study, and so the results will be discussed in relation to this recent model, as well as broader concepts arising in the interviews, such as trauma. Through this wider discussion, I will explore the applicability of this model, suggesting ways in which the model would need to be further developed to truly capture issues that we know are important in the context of parental caregiving. I will also end the chapter with an alternative suggestion of how we might conceptualise parental caregiving, based on the results of this thesis.

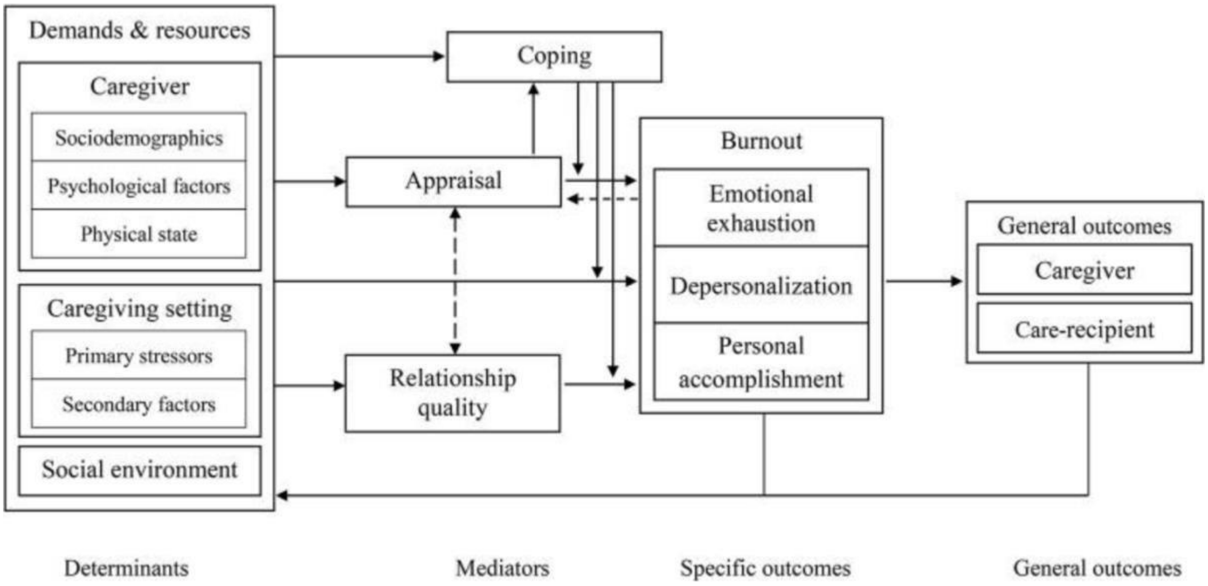


Figure 6; The Informal Integrative Caregiving Model (Gérain and Zech, 2019). Permission to reproduce the model as per the open access Creative Commons Attribution License (CC BY).

Below, I will revisit the findings of the mixed methods study, focussing on key findings, structured around the themes of the qualitative analysis alongside relevant findings from the survey. I will explore how my findings relate to concepts within the caregiving model as well as concepts drawn in from the broader literature. It is important to note that the themes often overlap, and so subsequent explorations of how each of these themes relate to existing knowledge will also overlap.

6.2 Fathers' experiences of precarity

The first theme of the thematic analysis highlighted the precarity experienced by fathers in navigating their daily responsibilities. Precarity has been defined as "a state of being, defined by its insecurity and vulnerability" (Buchanan, 2018). It is often used as a rather inflexible framework for understanding the impact of insecure labour conditions (Choonara et al.,2022). However, its application more generally allows us to explore and contextualise broader uncertainties, capturing lived experiences of precarity, as opposed to precarity as a social condition (Lain et al., 2019). Furthermore, this allows for us to examine how various markers of precarity intersect, as well as the impact of cumulative uncertainties (Lain et al., 2019). Precarity is increasingly used in health research, particularly in understanding illness experiences and uncertainties of older adults and their caregivers (Hillman et al., 2023, Portacolone et al., 2019). The theme of precarity in this study, arose from the sense of uncertainty, vulnerability, and fragility in fathers' accounts of their everyday lives and routines. This study is unique in that it explores broad and cumulative impacts of uncertainty on the lives of fathers of children with a life-limiting condition. It illustrates the uncertainties associated with their child's condition and how this creates uncertainties and fragility in their working lives and beyond. It also considers the uncertainties associated with obtaining appropriate care and support for their child, and fear of such being removed. There is significant tension in how fathers enact their various roles, related to this sense of uncertainty. Below, I discuss various aspects of, and contributors to, precarity as it relates to fathers; their lived experiences of uncertainty *alongside* the inherent unpredictability of their child's condition. I will also discuss these issues in relation to the caregiving model and broader literature where appropriate, but each contribute to this broader sense of precarity. Later in this chapter I will also suggest how the concept of precarity may be utilized in future studies surrounding the experiences of parents.

6.2.1 Role overload and the role of the father

This study has been able to explore the impact of employment status on the experiences, health, and wellbeing of fathers. Of particular interest in this respect, is the role **conflict** described by fathers in chapter 2 (systematic review), and the role **overload** and exhaustion described by fathers in chapter 5, usually related to the combination of their employment and caregiving roles. Over half of the fathers interviewed were in full-time employment alongside their caregiving responsibilities. Equally, the results of the broader survey demonstrate the nature of fathers' roles i.e., caregiving responsibilities alongside employment, with 26 fathers having made changes to their employment following their child's diagnosis.

Role overload has been defined as "a person's lack of resource to continue to fulfil multiple roles simultaneously" (Creary and Gordon, 2016) and "too many role demands given the time available to satisfy them" (Hecht, 2001). This is conceptually slightly different to the concept of role **conflict** identified in the review in chapter 2; here fathers' roles at work and at home were incompatible, with one often impacting on their ability to carry out the other (Creary and Gordon, 2016). These concepts are particularly relevant to our understanding of strain and burnout, as per the caregiving model (Gérain and Zech, 2019). Role conflict and overload are both dimensions of ***caregiver burden.*** There are several criticisms associated with the use of this term. It is conceptually ambiguous, incorporating the multi-dimensional consequences of caregiving including physical, social, financial, emotional, and psychological impacts. Furthermore, it has negative connotations that may mis-represent parents' actual experiences of caregiving for their child; burden is not a term frequently used by these parents. For the purposes of this study, and in line with how it has been more recently conceptualised by Gérain and Zech (2019), and in the broader caregiving literature, **caregiver burden** is taken to be an individuals' appraisal of the stressors associated with caregiving, i.e. as a mediator between demands/stressors and caregiver outcomes; it is "the subjective assessment of the stress that the helping situation can represent" i.e. part of **appraisal** in the model (Gérain and Zech, 2019).

Role theory (Georgas, 2004) centralises the concepts of overload and conflict, and may help to further elucidate the impact of caregiving on fathers. The theory has been used in this context as a framework for understanding burden, that is, how an individuals' perceptions, expectations and negotiations related to their multiple roles influence outcomes related to the demands/stressors of caregiving (Koch et al., 2021). Role development is termed '*role negotiation*', highlighting that roles are approached by individuals in ways that work for them and others within the context of that role (Major, 2003). If individuals are overwhelmed with demands, alongside a lack of resource, they may exhibit "unbalanced coping strategies" and develop mental health difficulties such as anxiety, depression, exhaustion, and subsequent burnout (Mullen et al., 2008, Major, 2003). The process of role negotiation is iterative, allowing for renegotiation when circumstances change, such as when a child is diagnosed with a life-limiting condition. We see in the results of the quantitative survey that many fathers made changes to their employment in order to provide care for their child i.e., will have required the renegotiation of roles. We also see in the interviews that fathers' multiple roles were inextricably linked, with each affecting the way in which they carried out another. These issues demonstrate the importance of understanding intersectional uncertainties in our understanding of precarity. Further markers of precarity are explored below, but a comment surrounding the impact of household precarity is warranted here. The links between fathers' roles, and indeed experiences of role overload, are particularly relevant to the discussion on precarity. Whilst fathers' jobs may be secure, uncertainties at home, including those related to change, may influence the way they perceive their working lives. Wider research frames this as an undermining of 'ontological security' (Lain et al., 2019).

How these role negotiations manifest in contemporary parenthood, and indeed fatherhood, is not fully understood. At a theoretical level, there are significant tensions in how fatherhood itself is conceptualised (Johansson, 2023). Contemporary theories recognise a reduction in gender-based role enactment at an individual level, and many families *do* strive for more equal role divisions, but inevitable social, psychological, and cultural influences add complexities to the levels at which this is possible. This is relevant because the **role overload** described above seems to be entirely reflective of the contemporary expectation that fathers be more involved with their children, and at home, alongside the enduring expectation that they should also provide financially for their families, creating difficulties in role negotiation (Johansson, 2023). The multidimensional nature of the transition to fatherhood requires role negotiation alongside individual internal working models, familial and societal and cultural expectations, as well as the individual characteristics of the child (Deave and Johnson, 2008). Recognising and emphasising these influences in the context of fathers of children with a life-limiting condition is relevant to understanding why these fathers may experience difficulties in relation to adapting to what can be rather ambiguous roles.

Findings surrounding fathers' employment and caregiving roles suggest that, although useful in some contexts, terms like 'primary' caregiver are less useful in describing families' caregiving arrangements and potential support needs. In the survey, fathers were asked explicitly about their roles in these terms i.e. ‘primary’ or ‘secondary’ caregiver or as having shared caregiving responsibilities with their partners. ‘Primary caregiver’ is broadly understood in the literature as an individual who is providing the majority of care to the recipient (Thomson et al., 2023) but it is possible that there were various interpretations of this term given that many of the fathers who identified as primary caregivers also worked full-time. What was interesting at the outset of this PhD was fathers' experiences alongside how *they chose* to define their roles as caregivers; how many of them considered themselves to be primary caregivers, and whether their experiences differed on this basis, which would be explored and elaborated upon based on how they described their family set up in interview. Of course, there were limitations to the dataset based on sample size, as discussed elsewhere, which meant that opportunities to explore this variable quantitatively were not possible, though I have to question what value this would have added given the point about various interpretations of primary caregiving above. Equally, in interviewfew fathers talked about their families' care set-ups using terms like 'primary' and 'secondary' caregiverThey described their caregiving responsibilities in the context of *family* caregiving, dividing responsibilities based on what worked for them and as mentioned, often worked *alongside* providing care for their child and is again reflected in the way in which the caregiving and employment questions were answered in the survey. Understanding the unique strains associated with full-time caregiving vs. full-time work *is* important but as roles continue to evolve there needs to be the recognition of nuance within family dynamics where terms like primary and secondary do not tell us much about the nature of responsibilities and may undermine caregiving contributions of the 'other' parent. Gérain and Zech (2019) highlight the importance of caregivers' socio-cultural environments, highlighted as a distinct demand/stressor. They reiterate the importance of understanding "the mechanisms by which culture affects caregiver stress and burnout", which is particularly relevant here when we consider fathers' experiences of work. Understanding these characteristics and the complexity of fathers' roles within the continuous evolution of family dynamics, will allow greater understanding of potential family support needs. All of this provides an opportunity for us to consider how we should define caregiving in this population; important not only for how we measure particular outcomes, but important within broader issues such as how parents perceive the relevance of research studies, as well as subsequent support and interventions.

A key aim of this study was to understand positive aspects of caregiving, including aspects of fathers' experiences that allow them to better cope with difficult experiences. Existing research tell us that being in employment may help fathers to cope with challenges at home, evident in the review in chapter 2. More generally, we see that the social connections available to caregivers at work, may mitigate the impact of stress on their health and wellbeing (Kokurcan et al., 2015). In the context of role theory this may be because they are able to retain a source of self-identity, and access a sense of respite from the often overwhelming and intense nature of caregiving, particularly if resources such as supportive and understanding employers are able to offer support (Major, 2003). Occupying multiple roles has been shown to improve wellbeing, however this was not reflected in the primary study. Explanations for this difference may be related to the COVID-19 pandemic, increased homeworking, less contact with colleagues, and less separation between work and home life, potentially also leading to the role overload described by fathers. This is important as it reflects potential contemporary changes to the consequences of upholding multiple roles, in the context of changing parental role and role ambiguity.

6.2.2 Unstable and inconsistent care systems

As well as a broad range of influences that impact how fathers perceive and cope with their roles, including their roles as caregivers, of particular importance is the practical support that they receive to support care for their child. The sense of role overload described by fathers, and resultant physical and mental exhaustion, was highlighted in relation to the support, or lack of support, that was available to them in helping to care for their child. The inequitable nature of specialist paediatric palliative care services is covered in chapter 1, though I think it is important to reiterate that not all families that would benefit from such services have access to them (Mitchell et al., 2019a). Alongside this are the increasing strains on other services, and hence variable levels of involvement in the delivery of palliative care for children, such as community nursing teams and GP's (Mitchell et al., 2021).

In this study, unstable and inconsistent means of support added to the precarity experienced by fathers in their daily lives; this spanned support provided by a range of agencies including health and social care. Furthermore, it seems that the impacts of the precarious nature of care arrangements were heightened during the COVID-19 pandemic, during which support like respite and overnight care were withdrawn, often with immediate effect, leaving many families to provide 24-hour care for their child with extremely limited information and support. Furthermore, restrictions meant that families had to isolate indefinitely (Scott et al., 2022), creating uncertainties about the future. Although it was not a key aim of this study, the timing of the interviews meant that such restrictions were still very relevant and prominent in the accounts of fathers. Consistent with research specifically exploring the impact of the pandemic on paediatric palliative care (Scott et al., 2022), the removal or delay of support, including means of psychological support for parents, alongside the withdrawal of certain care for their children, left fathers with a heightened sense of loneliness and vulnerability. Alongside the social isolation experienced by these families, were the financial, emotional, and psychological challenges. Families of children with complex needs are already disproportionately affected by socioeconomic challenges, including a reduction in working hours and housing insecurity (Mitchell, 2021).

The unstable care provisions that added to the existing precarity experienced by fathers is important to capture. There is inherent unpredictability and vulnerability when a child is diagnosed with a life-limiting condition, in relation to the often-unstable nature of their symptoms and condition. Literature touching upon how these fathers cope with uncertainties related to their child's diagnosis spans almost 30 years (Sterken, 1996, Mu et al., 2002, Hovey, 2005, Fortune et al., 2024). These uncertainties and the impact of such will be explored in further detail later in this chapter. Key to highlight here, and relevant to our understanding of precarity, is what happens when these uncertainties intersect with those created by fragile support systems. This in part touches upon communication difficulties with professionals, though is also related to the lack of control experienced by parents who feel that services are not adequately addressing the needs of their child. Furthermore, as highlighted during the COVID-19 pandemic, the withdrawal or failure to deliver certain types of care can mean that families are less able to cope with the uncertainties related to their child's condition. Again, these issues are not only important for recognising the support that parents need to provide care for their child, but the support that they need in relation to their own health and wellbeing. This withdrawal of support is important when we consider the impact of overnight caregiving, and therefore lack of sleep in caregivers, and the detrimental impacts that this can have on physical and mental health (Byun et al., 2016). Fathers in this study had worse sleep scores (PROMIS sleep disturbance (Hanish et al., 2017)), than those of the general population (Klapproth et al., 2022). Lack of sleep in this study was indicative of both a practical lack of support and fathers having trouble sleeping as a result of stress.

Importantly, research suggests that negative experiences as a caregiver, i.e. being abandoned by services/poor relationships with professionals, not only have a direct impact on the wellbeing of caregivers but subsequently means that they are less likely to participate in health promoting behaviours, compounding the effects of inadequate support (Tough et al., 2020). Existing research already demonstrates the need for better integration of the palliative care approach into existing healthcare structures, alongside improved coordination between services, supporting children and their families (Mitchell et al., 2019a, Mitchell et al., 2021). The frustrations felt by fathers in relation to this echo those captured in the broader literature, and indeed those of mothers in chapter 3.

6.2.3 Precarious relationships

Precarity was seen in fathers' relationships, including with friends, extended family and colleagues. Gérain and Zech (2019) highlight the breakdown of social relationships as being key contributor to burnout. The interviews with fathers, although not focused on burnout, did highlight the negative implications of the loss or breakdown of relationships with others, as well as changes to their relationship with their partner. The need for the renegotiation of roles helps to explain some of this, though the impact of relationship breakdowns, and subsequent caregiver isolation, in fathers, warrants further attention. Given the vital role that social connections play in the health and wellbeing of caregivers (Vasileiou et al., 2017), research has started to explore the impact of social isolation on informal caregivers more closely, though such studies are mainly focused on caregivers of adults (Hajek et al., 2021, Lee et al., 2021). Accessing support groups seems to improve the well-being of caregivers providing them with a better understanding of their child's condition, alongside feelings of hope and the realisation that they are not alone in their experiences, which in turn has been shown to help them to better cope with their child's condition and their caregiving roles (Koch and Jones, 2018, Bally et al., 2020). However, participants in this study, highlight the difficulties associated with finding support groups, particularly due to a lack of time. Furthermore, support groups for fathers were particularly lacking. These issues were heightened during the COVID-19 pandemic, though were not attributed to it directly.

Relationships with professionals could also be difficult for fathers. As discussed above, the multidimensional roles associated with caregiving can come with the need for greater levels of negotiation than those associated with 'regular' parenting. Alongside their roles as parents, often to healthy children alongside their child with a life-limiting condition, parents must provide extensive medical and personal care, coordinate all aspects of care and support for their child including busy appointment schedules, and advocate for them in multiple settings

(Koch et al., 2021). This means that parents must form relationships with a broad range of healthcare and support professionals, which requires negotiation (Brady et al., 2020). Indeed, this becomes even more pertinent in the case of fathers (rather than mothers), who report significant challenges in their relationships and communication with healthcare professionals (Polita et al., 2018, Ogourtsova et al., 2021).

Reflected in the review in chapter 2, we see that difficult interactions with professionals could reinforce fathers' perceptions around their roles in their involvement in their child's care, as well as how their own emotional needs could be met. In the primary study (chapter 5), a lack of recognition of fathers as caregivers, mainly in the context of their own support needs, created tensions in how fathers interacted with healthcare professionals more broadly. Existing research demonstrates the tensions that can exist with healthcare professionals when there is a lack of role clarity, particularly when parents are heavily involved in providing medical care for their child, including the monitoring of, and administering medication for, their symptoms and condition, and in which they therefore become experts (Brady et al., 2020). There often needs to be a greater recognition of this parental expertise and how this may shape their expectations and interactions with healthcare professionals (Brady et al., 2020). Negotiating these roles with healthcare professionals also has implications for shared decision-making, which we know can be important to parents (Corlett and Twycross, 2006). Further exploration of how these interactions shape fathers' functioning is so far limited, and research is warranted to understand how professionals can support fathers to enact their roles in ways that work for them and their families. Again, these issues are not only important for recognising the support that parents need to provide care for their child, but the support that they need in relation to their own health and wellbeing. Importantly, research suggests that negative experiences as a caregiver, i.e. being abandoned by services or ignored by professionals, not only has a direct impact on the wellbeing of caregivers but subsequently means that they are less likely to participate in health promoting behaviours, compounding the effect of inadequate support (Tough et al., 2020).

6.3 Feeling understood in the context of health and wellbeing

There is increasing interest in men's engagement in health service utilisation, particularly due to high rates of male suicide (Stiawa et al., 2020). With the added dimension of being a caregiver comes important factors that we must consider in this understanding of men's mental health. A key question raised by the findings of this study surrounds the type of support that would be beneficial for fathers in the context of their own health. The results of the survey demonstrated that fathers' main health concerns related to their mental health. There were also some slight problems reported in relation to pain and discomfort, mostly highlighted as back pain in the free text response and in the interviews. However, the EQ-5D and EQ-VAS scores were slightly higher (indicating better functioning) than those of the general population which sits in contrast to other studies exploring the health-related quality of life of parent caregivers (Poley et al., 2012, Arora et al., 2020). Such scores are also not reflective of how most fathers discussed their mental health and wellbeing in the interviews. This may relate to the inadequacy of the generic quality of life measure scoring, as it is heavily focused on physical health (Connell et al., 2014). There are also restricted inferences to make due to sample size limitations. Such findings do, however, reiterate the need for nuanced explorations of health, that are able account for issues that are important to fathers. Specifically, how do we capture the important manifestations of stress as experienced by fathers in this context? Bringing in the concept of burnout alongside these more general outcomes (Gérain and Zech, 2019) may indeed be useful, particularly when we consider fathers' experiences of emotional and physical exhaustion, feeling like they were at capacity, and not being able to attend to other aspects of their lives. Also, highly relevant here are the caregiving appraisals in the survey, through which fathers reported high levels of strain and distress. Specifically, the items within each of these domains cover issues such as feeling tired and run down, loss of social life, isolation, worry, anxiousness, guilt, and depression, alongside positive domains, providing a good overview of the appraisals of domains relevant to caregivers specifically (Cooper et al., 2006) and are entirely reflective of the experiences described by fathers in the interviews.

In the present study fathers' multiple role negotiations, alongside a lack of practical support, and having little time to do anything other than caregiving led to role overload, exhaustion, and a sense of vulnerability. These findings are consistent with those in the broader caregiving literature, and indeed with the review in chapter 2. However, the multi-dimensional and pervasive sense of precarity in fathers' accounts is not well reflected in existing models. This relates to the uncertainty that fathers described as a determinant, as an outcome, and as how they made sense of their experiences. The inherent uncertainties associated with their child's condition were compounded by uncertainties related to a lack of information, support, equipment, and care provision, alongside the uncertainties that this created for fathers in other aspects of their lives including employment and relationships, related to the future, both objectively and experientially. Uncertainty has long been established as an important construct in parental caregiving (Stewart and Mishel, 2000), but understanding this as a more ubiquitous construct in the experiences of this population may help us to understand the types of support, and changes to practice, needed.

The interviews highlighted differences in how fathers sought support for their mental health. There was some avoidance to fathers' support seeking, though there was not a clear distinction between for whom avoiding discussing mental distress was helpful and for whom it was a detrimental avoidance strategy; important when we consider the strong association between men who adhere to traditional male stereotypes and negative mental health outcomes (Eggenberger et al., 2023). This avoidance/delay is explored further below, through considering the importance of trajectories. It is well established that men are less likely than women to present to healthcare providers with mental health concerns, and we also know that the metrics with which we assess psychological distress are in themselves potentially unsuitable for reliably identifying distress in men (Martin et al., 2013). All of this is conducive to an environment in which fathers' mental health concerns may be underrepresented and overlooked, which is particularly evident through fathers' accounts of treatment by healthcare professionals involved in their child's care. Through the review in chapter 2, we saw that fathers' interactions with these HCP's could reinforce their assumptions about gender roles and support, and through challenging role negotiations identified in the primary study, we see how interactions with these professionals can influence fathers' own health seeking behaviours. However, the way in which fathers discussed their reluctance to seek support, fears around being misinterpreted, and a general lack of appropriate support, was similar to the perspectives of the mothers in chapter 3, suggestive of the need for improvements in addressing the health needs of caregivers regardless of gender, as well as a deeper look at the evidence suggesting that mothers are more likely to experience adverse health outcomes than fathers (Liu et al., 2023). Furthermore, one aspect of health that the caregiving model does not account for is the impact of poor health on the risk of repeated ill health (McDonald et al., 1997)

The high levels of strain and distress reported by the fathers in this study, are comparable to those as reported in a large sample of caregivers (88% mothers) (Collins, 2020) further supporting this argument. This is further elaborated on in section 6.5 where I discuss key similarities and differences in the accounts of mothers and fathers.

6.4 Trajectory of child's illness and importance of temporality

Due to the heterogeneity of life-limiting conditions in children, there are various routes to diagnosis, some longer than others if, for example, genetic testing is required (Hoell et al.,2019) Hurley et al. (2021) explored parental experiences of their child's diagnosis of a non-malignant life-limiting condition. Although only 3 of 23 participants were fathers, many of the experiences and emotions described by parents were reflective of the experiences identified in this thesis, both in the review in chapter 2 and those described in the interviews with fathers in chapter 5. In all instances, parents recalled the moment of diagnosis with vividness, describing powerful emotions. Some fathers were particularly troubled by the way in which HCP's had delivered the news of their child's diagnosis. Poor delivery of diagnosis was common and is widely documented in the field of paediatric palliative care, highlighting multiple issues surrounding poor communication, information provision and a lack of compassion from professionals (Hurley et al., 2021, Ekberg, 2018, Hill and Coyne, 2012). How this affects parents, and in this case fathers, is important for their own wellbeing, including how they negotiate their own role, and how they subsequently interact with services in the context of their child's care. There is evidence to suggest that exposure to a traumatic event may interfere with a caregivers ability to engage in similar events in the future; i.e. the impact of a traumatic diagnosis could be to avoid information seeking behaviour related to health in the future (Sun et al., 2022).

6.4.1 Repeated traumatic experiences

In the findings around trauma, in the review and primary study, fathers described multiple and repeated traumatic experiences, including the use of the phrase "trauma-on-trauma" meaning they had little opportunity to manage past traumas given current caregiving responsibilities and subsequent distressing experiences as well as many fathers being in a state of constant hypervigilance. The Diagnostic and Statistical Manual of Mental Disorders Fifth Edition (DSM-5) considers "a medical catastrophe concerning ones child" to be a traumatic event, and we do know that caregivers of children with life-limiting or life-threatening conditions are at an increased risk of post-traumatic stress disorder compared to other parents (van Warmerdam et al., 2019, Conijn et al., 2022). Clusters of symptoms of PTSD include intrusions, avoidance and negative alterations of mood and cognitions (Conijn et al., 2022). Although not all parents will experience these symptoms in a way that warrants a diagnosis of PTSD, one such way that parents' symptoms have been described is through the Integrative Model of Paediatric Medical Traumatic Stress (PMTS) (Kazak et al., 2005). This model considers PMTS to be "a set of psychological and physiological responses of children and their families to pain, injury, serious illness, medical procedures, and invasive or frightening treatment experiences" (Kazak et al., 2005), which is particularly useful for identifying potential support without the need of a psychiatric diagnosis. This is pertinent in the consideration of fathers' experiences in this study, through which we understand that few fathers had a formal diagnosis of PTSD but described their experiences as *traumatic*, and the effect of this as ongoing and unresolved distress, compounded by further traumatic experiences and uncertainty. This is also particularly relevant when we consider parental perspectives of pathologising distress.

The majority of what we understand about trauma and parental mental health comes from literature surrounding paediatric oncology populations (including those who have recovered) (Feudtner et al., 2021), meaning that we know little about how this trauma may manifest in families of children with life-limiting conditions with very different trajectories and treatment profiles. If we also consider traumatic stress in relation to appraisal, and that parents' perceptions of their child's condition and threat to life are hugely important in understanding the stress response and subsequent risk of PTSS or PTSD (Lindahl Norberg et al., 2012), we can see why the traumatic experiences described by fathers in this study were very much related to the fragility and uncertainty of their child's condition and prognosis. Many fathers found it difficult to make a reasonable assessment of the severity of their child's current health state given either a lack of information/support or because of the inherent instability of their child's condition, which also contributes to how fathers make sense of their own roles. Information and support, both informational and emotional, was particularly lacking during, and in the time immediately following their child's diagnosis, but we see this repeated at various timepoints throughout their child's illness. Recent research highlights the need for trauma-informed palliative care (de Groot et al., 2023), though current evidence surrounding suitable approaches are limited. Fathers' narratives around traumatic experiences are certainly indicative of the need for support that is able to address such distress, as well as minimise the traumatic effects of future experiences.

As highlighted, the conceptual model of caregiving (Gérain and Zech, 2019) centralises the importance of caregivers' appraisals of stressors on the development of tridimensional burnout (emotional exhaustion, depersonalisation, and personal accomplishment). Through the results of this study, we see that emotional exhaustion is extremely relevant in understanding the experiences of fathers. In parallel to this is 'personal accomplishment', encompassing the positive effects of caregiving. One of the aims of this study was to explore the positive dimensions of caregiving and one concept that has emerged through the exploration of trauma, is the concept of posttraumatic growth (PTG). 'Posttraumatic growth' was introduced by Tedeschi et al. (1998) to describe positive psychological changes following on from a traumatic event. In the context of caregiving, posttraumatic growth has been suggested as reducing or preventing the burden experienced by caregivers in their roles (i.e. appraisal of stressors). However, most research surrounding PTG has been carried out in adult settings, and in childhood injury (Ni et al., 2023), and it is not surprising that we know little about PTG in the context of life-limiting illness in children, given the dearth of research on trauma in this area more generally. Understanding means by which fathers, and families, cope well with their child's illness is key in further understanding what type of support helps or what type of support can enrich helpful coping mechanisms. This could also help to further explore whether personal accomplishment is a suitable metric with which to assess the outcomes of caregiving. The diagnosis of a life-limiting condition presents families with challenges in all aspects of their lives. The fathers in this study described ongoing losses, having to re-shape their expectations of the future; both the sense of loss they experienced related to their child's future as well as what they had envisioned for themselves as individuals and as parents i.e. the role negotiations crucial to fathers' adapting to change. More research is needed to assess how traumatic experiences shape fathers' broader experiences and how the effects of such experiences can be minimised or promote more positive experiences. Understanding how best to conceptualise this in the context of parental distress is also important i.e. distress resulting from the everyday worry that their child will die, as opposed to those acute periods of distress related to diagnosis, prognosis, and critical periods of illness.

6.4.2 Relationship with their child

One part of fathers' narratives surrounding loss, was the change in the relationship with their child. This is discussed under the trajectory theme, as it related to changes in this relationship, often as a result of changes to their child's condition. For others it was comparisons to siblings, or to their own expectations of the father-child relationship. This is touched upon in the discussion surrounding role theory, but it is also relevant in the context of trajectory and feelings about the future. Changes to the nature and strength of relationships are often central to caregiving research. Such changes form a central mediating factor in the model, because it is well known that fluctuations in caregiver and care recipient relationships impact caregivers' appraisals of their caregiving role and associated stressors, subsequently impacting their wellbeing (Gérain and Zech, 2019); poorer quality relationships have been linked to role overload.

However, it is important to note that the impact of this relationship has so far mostly been understood in the context of caregivers of adults, as opposed to in parent-child dyads. This is also the case for "depersonalization" in the tridimensional concept of burnout, defined as "the detached response in the relationship to the person being cared for. Taken to the extreme, this can go as far as the reification..." (Gérain and Zech, 2019). Again, there has there been little research into this in informal contexts, but even less so in the parent-child relationship. We see that the transition to caregiving roles is experienced differently within different types of relationships, alongside different motivations for caregiving and role negotiations. Although the caregiving model is clearly applicable to various caregiving contexts, how relationships have been conceptualised within the model may not be, and we may be missing important aspects of parent-child relationships and how they manifest in caregiver outcomes and in clinical practice.

Attachment theory tells us that the thoughts, feelings and behaviour that underpin child-parent relationships operate with the purpose of protecting the child (Bowlby, 1969). When a child is unwell, parents are required to re-work their internal models of their child and themselves as parents, so called *resolution,* as a means of managing the reality of their child's illness; a form of coping, with similarities to classic emotional regulation and problem-solving strategies (Lazarus and Folkman, 1984, Lazarus, 1966, Lazarus, 1996). This is extremely important within the trajectory of illness, as the process of *resolution* can take time, with parents living in the paradox of knowing their child will die whilst looking for curative or life-extending treatments. Research has shown that parents view their responsibility as "decision makers for their child, as they both perceived and enacted it" and "to leave no stone unturned" (Bluebond-Langner et al., 2007). Related to this is the way in which parents have been shown to reshape their expectations and plans for the future, based on their child's transitions to new stages in the illness trajectory (Bluebond-Langner et al., 2017).

The concept of trauma-on-trauma and the impact of adverse events on fathers' future experiences and perceptions of their child's condition and healthcare is very much related to concepts discussed within attachment theory. Those parents with high distress and extended periods of grief are more likely to suffer longer-term adverse outcomes, particularly in their mental health, and when we consider that parents want to fulfil a protective role at all costs, we can understand how traumatic it may be when they feel unable to have done so (Nicholas et al., 2016). Fathers must be well supported through potentially traumatic experiences and models of caregiving need to recognise the unique complexities within the parent-child relationship that mediate caregiver outcomes. Using concepts from attachment theory is one way to do this, even if it is just the recognition of individual parental characteristics and how they influence the parent-child relationship. Despite its inclusion in the original model, recent evidence suggests that relationship quality has a relatively low predictive value in relation to burnout (Gérain and Zech, 2022), further suggestive of the need to reconceptualise this aspect, taking account of the losses associated with a child's condition, not only impacting fathers in the sense of being a caregiver, but as a parent. Equally, the way these changes manifest in relation to burnout are perhaps not best recognised as depersonalisation because the concept has not been tested in populations outside of professional burnout, and certainly not in parent-child dyads, as discussed. We also know that such negatively loaded words can fail to resonate with parents, who do not want to stigmatise their child or the care that they require. Regardless of whether these negative appraisals are captured alongside more positive evaluations, it is seemingly inappropriate to use language that is in conflict with the language that parents would naturally use to describe their experiences (Koch and Jones, 2018).

6.4.3 Anticipating suffering

Fathers described their future worries surrounding their child's suffering, including fears about their death. One way that this has been conceptualised is in terms of anticipatory grief. Anticipatory grief has been described as "phases of mourning, coping, interaction, planning, and psychological reorganization in response to the imminent loss of a loved one" (Najafi et al., 2022). However, when we consider that children may live with a life-limiting condition for many years, we understand that the experience of anticipatory grief may extend over a similarly long period of time. Singer et al. (2022) proposed that *pre-death grief,* made up of anticipatory grief and illness-related grief is a useful delineation; anticipatory grief is focused upon fears about future loss, including death, whereas illness-related grief encapsulates all present day losses related to the illness (Singer et al., 2022). This is very much in line with the way in which the fathers in this study distinguished various forms of distress, both current losses and re-shaping expectations (as above) alongside this anticipatory grief, focused on their child's death. Distinguishing these forms grief is helpful as a means of identifying targets for support and interventions, and also useful in delineating various types of trauma as explored above. For children and families who have access to paediatric palliative care services, there will often be grief support available long before the child's death. However, as discussed, not all families have access to such support.

As previously discussed, the term 'life-limiting condition' covers such a vast array of conditions, severity of symptoms and needs, that it is not always possible to predict the trajectory or prognosis for individual children and many will live into and beyond their teenage years, some into adulthood. Whilst some of the children in this study had progressive illnesses, such as Duchenne Muscular Dystrophy, many had conditions with less clearly understood trajectories. Furthermore, many of these children were at risk of dying not from the condition itself, but from vulnerabilities to other illnesses as a result of the condition, creating immense uncertainty for families. How we understand the temporal aspects of parental experience is important for understanding the timing of support for fathers and the broader family unit. Understanding how this might vary for fathers of children with progressive illnesses compared to other types of life-limiting conditions is also important in thinking about appropriate support or interventions. Furthermore, individual characteristics and cultural expectations must be considered in our understanding of anxieties around death.

A major limitation of this study was the lack of cultural and ethnic diversity, in the broader quantitative sample, as well as the qualitative sample. This is particularly important because the sample does not sufficiently represent the ethnicities of those most affected by paediatric life-limiting conditions.

6.5 Comparing findings with the experiences of mothers

This thesis provides the opportunity to compare the experiences of mothers (chapter 3) and fathers (chapter 5) of children with a life-limiting condition, more directly than the existing literature allows for. Understanding the unique experiences of fathers, independently from those of mothers, is of course important, and the main body of this thesis allows for this. However, not only does the comparison of experiences offer deeper insight into caregiving but offers the chance to explore the effects of contemporary parenthood and changing roles in relation to other important contextual factors, including gender and employment, to how we understand these experiences. This is important for the development of effective support/interventions for this population of caregivers as well as how future research is framed and approached.

6.5.1 Psychological interventions for caregivers

Mothers and fathers both discussed their experiences of poor mental health and psychological challenges. We know from the existing literature on mothers (Fraser et al., 2021), and emerging data on fathers (Liu et al., 2023), that the prevalence of anxiety and depression is higher in parents of children with a life-limiting condition than other parents. These outcomes have been heavily studied in the context of caregiving/family caregiving more generally, and so it is unsurprising that they feature in the accounts of both mothers and fathers of children with a life-limiting condition. However, a key similarity in parents' accounts were their frustrations surrounding clinicians seemingly pathologising their reactions to their child's illness, and subsequently providing inappropriate or ineffective support or treatment, highlighting the need for a more nuanced understanding of the mental health of these parents. Both mothers and fathers had found short course psychological therapies, such as CBT, to be inappropriate for them as caregivers. This was attributed to several reasons including lack of practitioner understanding of what caregiving for a medically complex child entailed, frustrations surrounding it being suggested that their reactions to their child's diagnosis and condition were abnormal, and the inherent nature of short-course therapies being able to offer longer-term and more sustainable support. The last point was particularly prominent in fathers' accounts i.e., that more, or less, support may be needed at various points over the course of their child's illness. For mothers, this resistance to such therapies was more heavily related to their inability to address the underlying causes of their distress i.e. battles with services for support and equipment for their child. Fathers did touch upon these battles, though the presence and effects of such were not as prominent in their accounts as they were in mothers. This may be related to the hours during which parents are in contact with services (working hours during which employed fathers were likely to be working).

Although interventions based on cognitive models clearly have some benefits in the management of stress and appraisal, using therapies to address 'cognitive vulnerabilities' in these parents needs to be explored with greater consideration of their underlying experiences, and not just the presence of symptoms of anxiety/depression. Parents' experiences of CBT in particular, resonate with existing studies demonstrating its inability to resolve caregiver burden (an individuals' subjective assessment of the broader consequences of caregiving) (Hwang et al., 2022). Equally, in relation to the concerns of fathers, reviews tell us that there are few psychological interventions for caregivers that produce long-term benefits (Greenwood et al., 2018).

6.5.2 Experiences of trauma

As detailed in chapter 5, fathers spoke specifically about trauma. Some mothers did discuss their mental health experiences in relation to post-traumatic stress, but references to trauma specifically were less common than in the accounts of fathers. Importantly, mothers did describe their experiences of their child's diagnosis, with the same vividness as fathers, as well as using numerous words synonymous with trauma. What was unique about fathers' accounts was their focus on the impact of repeated traumas, and the impact of this over the extended course of their child's illness. The ranges and mean ages of the children in both parental samples were similar, giving both groups the opportunity to explore experiences over similar timeframes. Initially, the results seem to sit in conflict with what we know about traumatic stress i.e., women are much more likely to meet the threshold for clinically relevant

PTSD than men (Olff, 2017). However, research also suggests that despite this difference, women are *less* likely to report having experienced a trauma (Jingchu et al., 2017). However, it is important to distinguish between existing prevalence studies, and the impact of trauma described by parents here; little research currently exists that demonstrates gender differences in symptoms of PTSD, experiences of trauma, and the potential explanations of such in these parents (Carmassi et al., 2018). Furthermore, the underlying mechanisms of such are complex. Understanding parental perceptions of trauma, including the successful navigation of such may help to explain how, and why, these parents experience certain outcomes.

Gender differences in response to trauma can sometimes be understood through coping, a key aspect of the caregiving model. Women are more likely to use emotion-focused strategies, whereas men are more likely to use problem-focused strategies to deny or avoid stressors (Altınsoy and Aypay, 2023). When we consider fathers' focus on the trajectory of illness, and worries for the future, we see that there may be substantial conflict between their usual coping mechanisms and what is possible in the context of their child's illness. Using problem-based coping strategies alongside the overwhelming sense of precarity in all aspects of their lives, may have resulted in what they described as a lack of opportunity to process trauma and hence focussing on this in their accounts. However, as discussed, this is a complex issue warranting a greater understanding of coping processes over time in the context of childhood life-limiting illness.

Also relevant to our understanding of trauma, and coping, is the concept of resilience (Manzari et al., 2023). Resilience can sometimes help to explain how some parents avoid distress, alongside the objective demands of caregiving. Although this is another concept that is conceptually multi-faceted, it has broadly been defined as adapting in the face of difficulty and resisting the negative impacts of stress (Mezgebu et al., 2020). If we consider the results of the systematic review in chapter 2, we see fathers' accounts of adapting to the changes and uncertainty associated with their child's condition; the associated stresses became part of their everyday lives and 'the new normal'. This is similar to findings in the broader caregiving literature, particularly studies on mothers, that demonstrate the ability to navigate difficult and consuming care routines and fulfil responsibilities under challenging circumstances i.e., resilience (Baker and Claridge, 2023). However, as demonstrated through this study, there is a need for new perspectives on parental experiences of trauma, able to distinguish between various sources and manifestations of such. Understanding that parents have adapted to extensive and exhausting caregiving routines does not tell us much about how their own physical and psychological needs are being met, or how this will affect their response to difficult experiences in the future. Whilst coping and resilience may be useful concepts, they again need to be considered within the broader family experience including alongside role negotiations and expectations and over the course of illness.

6.5.3 The protective roles of mothers and fathers

The desire to protect other family members from distress was prominent in the accounts of both mothers and fathers. Many fathers attributed this directly to their perceptions of what it meant to be a father/partner. This is very much reflective of what I found in the review in chapter 2, through which fathers described this same sense of being the protector or 'the strong one' for their family. However, mothers also highlighted a need to be strong for others and to protect their partners from further psychological distress, which involved keeping their own emotional struggles to themselves. Others' perceptions, particularly those of HCP's, of how well they were coping, were also particularly important to mothers. In this study we have seen how family roles can be played out without attribution to traditional gender norms, but through what works for individual families. However, as part of this broader negotiation, the way in which mothers *and* fathers described managing and sharing their emotions was very much rooted in the prevailing sociocultural norms surrounding what it means to be a good mother or a good father. Much of this was related to role conflict and uncertainty for fathers, as explored earlier in this chapter. Maternal shame, although not well understood and even less so in mothers of children with complex needs, is associated with this sense of unwillingness to disclose emotions or to seek help (Sonnenburg and Miller, 2021). Shame is also strongly related to guilt, fears around 'failing' as a mother and developing a new sense of self (Law et al., 2021). Such concepts are extremely relevant not only to mothers but in our understanding of the importance of cultural narratives of parenthood are able to influence both mothers' and fathers' experiences and health behaviours. Such concepts are also relevant to the 'relationship quality' problem explored earlier.

6.5.4 How parents look after their own health

Mothers and fathers both described similar health concerns, including anxiety, depression, and back pain. The way in which these issues are perceived and addressed by healthcare professionals is touched on above. A key difference in the accounts of mothers and fathers was the way in which they addressed their own mental and physical health concerns. There were several reasons why mothers did not feel able to prioritise their own health needs, touched upon in chapter 3 and in the threads above, related to inappropriate offers of support, fears around how their concerns would be perceived, and wanting to protect others. Mothers also described being fearful that if they did become unwell, there would be no-one to care for their child. Another issue for mothers was that healthcare services, such as GP's, did not seem to have the capacity to address their needs alongside those of their child's, which were the priority. On the contrary, there was a greater sense in fathers' accounts that, should they need to, they could approach healthcare professionals about any specific health concerns they had. Despite this, fathers did describe instances in which they had put off seeking help because of unclear thresholds for support i.e., they were unsure as to whether their concerns did indeed warrant support or care. This meant that there could be similar delays in mothers *and* fathers receiving the care they needed, or care that could have helped them sooner. Both mothers and fathers felt more comfortable in seeking support through their local children's hospice than through other means. These were environments in which parents felt able to express their emotions, without fear of judgement from hospice staff. This is very much reflective of literature demonstrating the benefits of specialist palliative care services, including hospices, through which families' needs are addressed more holistically.

6.6 Precarity and caregiving

Throughout this chapter I have referred to aspects of the conceptual model of caregiving (Gerain and Zech, 2019) and described ways in which the model would need to be adapted to ensure that it captures important aspects of fathers’ experiences. Insufficiencies of the model were particularly evident in relation to the unpredictability of fathers’ experiences, the fluctuation of their child’s condition over time, unaddressed trauma, and aspects of the parent-child relationship. As mentioned, although caregiving models may be useful in capturing general experiences and outcomes, and were indeed useful in framing and exploring fathers’ experiences in this study, further work is required surrounding how we conceptualise parental caregiving in this population. As demonstrated through this thesis, precarity is an important and prevalent issue in parental accounts of caregiving. Elements of precarity can be seen across the data in each of the qualitative studies, cutting through individual themes within studies, and providing an important construct for how we might understand parental experiences and the impacts of such more clearly. Although it is through the fathers' data that the concept of precarity is particularly emphasised, mothers' accounts of uncertainty, a lack of information, feeling abandoned by services, a lack of tailored support, changes in relationships, career changes, financial instability and fears about the future are just several examples of what could be considered as markers or components of precarity (Llosa et al., 2024).

As mentioned, the impact of precarity has most often been studied in relation to workplace insecurity and a recent study has conceptualised the impact of a precarious life on health and wellbeing (Choonara et al., 2022, Llosa et al., 2024). Another recent, and particularly relevant, study has used precarity as a framework for understanding the experiences of caregivers of dementia patients, describing the framework as holding "within it both material and existential threats to personhood" (Hillman et al., 2023). The study describes and incorporates many of the same challenges experienced by the parent caregivers in this thesis, such as the fragility of care and support systems and the presence and impact of cumulative stressors. Importantly, the study also differentiates precarity associated with meeting care needs and the precarity inherent to the condition itself. This is an important distinction and should be considered not only in any future conceptualisations of precarity but in how we might mitigate the impact of such through support and interventions. The parents interviewed in the studies in this thesis emphasise this distinction, highlighting that much of the uncertainty they experience is related to absent, inconsistent, inappropriate, and untimely support, rather than their child's condition itself. In assessing and potentially adapting any frameworks of precarity, care should also be taken to ensure that the specific challenges or vulnerabilities faced by parent caregivers are included and that key experiences and concepts are not missed, as has been the case with adaptations of current caregiving models. This thesis goes part of the way in identifying some of these particular vulnerabilities, but as discussed below, the homogeneity of parental samples needs to be improved upon if we are to truly develop a meaningful framework to guide our understanding. Future studies should seek to recruit those living in areas of high deprivation and those from minority ethnic groups to inform a framework containing the various components of precarity relevant to this population of caregivers.

6.7 Strengths and limitations of thesis

Through this thesis, I have addressed a gap in the literature by exploring the experiences, health, and perspectives of caregiving of fathers of children with a life-limiting condition. The systematic review in chapter 2 found 32 papers from 30 studies that explored fathers' experiences. These studies had several limitations including an under-representation of UK based fathers of children with complex neurodisability. Furthermore, they did not directly address the health of fathers. Therefore, the primary study on fathers has first and foremost provided me with the opportunity to engage an under researched and poorly understood population to give rich and detailed accounts of their experiences, allowing for in-depth explorations of their perspectives. In addition to the findings relating specifically to fathers, through wider work with mothers I have been able to directly compare their experiences and support needs in the context of caregiving and their own health and wellbeing. In reference to the theoretical model of caregiving, I have been able to demonstrate the relevance of important aspects of the model, as well as how our understanding of parental caregiving could be better represented in this model and those like it.

The sample size for the fathers' study was smaller than expected. Although I suspected that the initial target for the survey was ambitious, the final sample size was still lower than I had anticipated it would be. This means that it was only feasible to conduct descriptive statistics on the final quantitative dataset, rather than more detailed associations and regression analyses. This also had implications on the nature of integration with the qualitative data, which would have perhaps provided deeper insight had there been more scope in the analysis of the quantitative data. However, the survey was still able to provide important contextual information about the fathers taking part in interview, as well as providing them with the opportunity to engage with the topics that would be discussed in the interviews. During the interviews that I conducted with mothers, I often found it difficult to encourage them to discuss their own health and wellbeing specifically. Although this was related to the findings, in that mothers' health was inextricably linked to their child's, I found it easier to encourage fathers to talk about their own health alongside their experiences of caregiving, which was surprising. This may be to do with my own development as an interviewer, but there is evidence to suggest that various forms of concept priming are useful in encouraging respondents to discuss them specifically (Vitale et al., 2008). Of course, this is all usually detailed in a participant information sheet, but through engaging with the survey questions, fathers were asked very specific questions relating to the impact of caregiving on their health, meaning that when it came to the interview, they had already had the chance to think about these concepts more than an information sheet would encourage. Despite the final sample size for the interviews also being smaller than the target, I was still able to collect rich and detailed information surrounding the health and caregiving experiences of fathers. Given the extended recruitment period for the fathers' study, alternative strategies may have been useful.

There was a particular lack of ethnic diversity in the samples of mothers and fathers, not only in the primary studies but also in the systematic review in chapter 2. Caregiving outcomes in parents from ethnic minorities are important to address in the next phase of this research given the prevalence of life-limiting conditions in ethnic minority groups. Furthermore, attention should be paid to including those from areas of highest deprivation where prevalence is also high (Fraser et al., 2021). This lack of representation is particularly important to address as we start to think about what support, services, and interventions might be appropriate for these families. Many of the parents in this thesis spoke highly of the support available to them through their local children’s hospice. However, despite the benefits of children’s hospices, we know that those in ethnic minority groups and those living in areas of high deprivation are less likely to use them than other groups (Pentaris et al., 2018).

Further work needs to be done surrounding recruitment strategies that encourage diverse participation, and that go beyond recruiting 'fathers'. In this study, there were several streams of recruitment, including social media, and letters, emails, and face-to-face recruitment at participant identification centres. Furthermore, there were participant identification centres at various locations across the UK. There was slight improved diversity in terms of household income, but intersections between ethnicity and deprivation are important to explore in this context, given the prevalence of conditions in these groups alongside associated disparities in palliative care provisions (Kaye et al., 2019).

6.8 Conclusions

Caregiving should be considered in the context of individual families and relationships, that includes the consideration of parental support needs regardless of employment status or hours spent caring for their child. As parenting roles continue to evolve, so will the tensions associated with how individual parents negotiate these roles. What is clear is that overload and strain are not just related to the accumulation parenting and other roles, but to these complex negotiations, with various parties, that fathers must undergo in the development of such roles. In the case of fathers of children with a life-limiting condition, this not only includes their partners, employers, friends, and families, but healthcare professionals involved in the care of their children. There are important considerations in how we understand family relationships in the context of childhood life-limiting illness, and nuances to the parent-child relationship that have so far not been accounted for in models of caregiving. Further work is required to develop a model that truly considers parent-child dyads, including in how the concept of burnout, mainly with regards to relationship quality, is conceptualised within the model. Although burnout seems to be a useful concept in terms of the physical and mental exhaustion that fathers described, the current model does not capture this fully. How fathers build relationships with, and negotiate roles and responsibilities with, numerous healthcare professionals involved in the care of their child also requires further attention, as does the impact of ongoing strain within such relationships. Furthermore, how these roles are negotiated, and renegotiated, in the face of multi-dimensional precarity, should be explored.

The precarity in fathers' accounts demonstrates the constant sense of insecurity and vulnerability that fathers experienced. This goes further than current understandings of caregiver uncertainty, as it explores the impact of cumulative uncertainties; the fragilities and unpredictability inherent to their child's condition and care, as well as those associated with the broader aspects of fathers' lives including relationships and employment. Although the first theme is centered around **precarity**, I have shown that precarity is a cross-cutting theme across the dataset. Subsequent themes also contain aspects of precarity whether in relation to relationships with professions, family members, and friends, or in relation to future plans, or the trajectory of child’s illness. How such manifests in terms of health and wellbeing needs to be considered, perhaps not as a set of determinants as per the caregiving model (stressors or demands), but as an intrinsic element of their experiences that spans demands, appraisal, specific and general outcomes. Understanding the impact of various forms of precarity, i.e. as a direct result of their child's condition or as a result of unstable support or care provision alongside the more existential precarities experienced by fathers also reiterates findings from the mothers' study, in that it is difficult to disentangle parental experiences from the needs of their child. Support tailored to these parents should account for this. There are important differences between parental distress because a child is not getting appropriate care and support, and distress because the parent is not being well supported themselves. Precarity is a useful and important concept for how we might understand these experiences and future work should explore models of precarity.

The cumulative nature of the distress experienced by fathers throughout the course of their child's illness was also captured by the concept of trauma-on-trauma. The way in which the literature currently conceptualises trauma in relation to childhood injury and illness is useful, though does not fully incorporate important considerations of temporality. Fathers' needs will fluctuate over the course of their child's illness, demonstrated across themes, but there are key timepoints for professionals to be mindful of in the trajectory of the child's illness and in the development of trauma-informed palliative care (de Groot et al., 2023). This has not yet been fully explored, particularly in terms of how fathers' past experiences, in relation to their child and their own health, shape future expectations and perspectives, as well as their ability to cope with stress.

Current mechanisms by which parent caregiver support needs can be identified and addressed do not sufficiently capture important aspects of their experiences. Mothers and fathers were both reluctant to address their own health needs, often due to the lack of availability or appropriateness of support options. Psychological interventions, often provided in short courses, were unable to account for the specific nature of caregiver experience including the unpredictability and parental health being intertwined with the needs of their child. Children's hospices were able to provide more appropriate support for parents, particularly because of their innate understanding in relation to the lives and challenges experienced by parents.

6.9 Implications for policy, practice, and research

Research highlighting the experiences and support needs of these parents is increasing but how this translates to policy and practice has so far been limited. Healthcare professionals, such as GP's and mental health practitioners, are not necessarily equipped to understand the complexities of this role and subsequent parental needs. Central to the experiences of parents are the tensions in making sure that their child and family are well cared for, alongside them managing their own health and wellbeing. Services need to work in ways that ensure all family members feel supported and that any assumptions relating to roles/impact of caregiving do not hinder this work. It is important that we can provide a more detailed understanding of access to and preferences for service provision, including specific barriers to access that ethnic minority groups and those living in areas of high deprivation may experience. We know that language and cultural barriers, as well as a lack of awareness of available support, may make up some of these barriers (Carers UK, 2010), and so it is important that we employ inclusive research methods and recruitment strategies to ensure that these voices are included. Not only do we need to further understand the issues faced by these groups, but we need to translate this into practical, feasible, and meaningful recommendations for the development of services and interventions. Cultural differences in role functioning and support seeking will be particularly important to understand, particularly when we are thinking about concepts such as 'family-centred' and 'hollistic' care and support, for which individuals may have varied understandings and/or priorities.

Concepts in the model of caregiver burnout (Gérain and Zech, 2019) are relevant to the experiences of fathers, but as explored at various points of this discussion, there are certain aspects of this model that require further investigation. The model does not account for the very pervasive sense of precarity in fathers' accounts, nor is it able to demonstrate how fathers navigate this in relation to the trajectory of their child's condition. How we understand the nature of parent-child relationships in the context of caregiving also requires further attention. Using precarity as a framework for these experiences may help to resolve some of these tensions but additional work is needed to include the experiences of a more diverse range of parents.

Parents were particularly well supported by children's hospices in this study, demonstrating the value of environments in which parents are; (a) able to receive support and treatment alongside that of their child; (b) given the space to talk openly about their experiences, with professionals and other parents, who have knowledge surrounding the nature and impacts of caregiving; (c) able to get practical support with caregiving, from professionals they trust. Relationships with HCP's matter; these relationships can help parents to cope with changes, negotiate their expectations and make decisions. Key to this is how access to children's hospices and other specialist palliative care providers can be improved for these families. However, as touched upon above, including a more diverse range of voices in understanding more about which services should support these families, should be a priority. Furthermore, other changes are needed to improve the coordination of other service that are outside the remit of children's hospices e.g. education and equipment. Services need to be capable of prioritising the needs of the child alongside *responding* to needs of parents. Given the reality of how services operate, the incorporation of better/appropriate support for fathers, and indeed parents generally, may not be straightforward.

One intervention that may be able to address some of the shortcomings of current support provision for parents, is the Carer Support Needs Assessment Tool intervention (CSNAT-I). This intervention has been shown to be effective for caregivers of adults with palliative care needs and has recently been assessed for its suitability for use with parents of children with a life-limiting condition, using the data presented in this thesis (Fisher et al., 2023). The evidence-based question-and-answer format of the tool sits within a five-stage intervention process, capable of addressing parents' own health and wellbeing needs alongside any support needed to help parents to care for their child. Further work is required to assess feasibility and acceptability of use, as well as to identify whether further adaptations are required in the case of other cultures or contexts, as highlighted and discussed above. However, The CSNAT-I is able to address the tensions surrounding how parents manage their own needs alongside those of their child. Related to this is the need for further research surrounding how we refer to parent carer/caregivers and the implications these terms have for how we understand the experiences of these parents. Framing their experiences, and indeed any interventions such as the CSNAT-I, around concepts related to caregiving may indeed be useful. However, as highlighted throughout this thesis, how parents define their own roles is not necessarily in line with how caregiving roles have been defined in the literature, particularly in terms of ‘primary’ and secondary’ caregiver. The tensions associated with parenthood and providing high levels of medical care are important to acknowledge and account for as well as how we might formally define caregiving in this context.

6.10 Summary

In this chapter, I have explored how findings in this thesis add new understanding to the experiences and health of fathers, as well as how they compare to the experiences of mothers. I have also described implications for practice and research. There is currently limited support able to address the unique and fluctuating support needs of fathers, which are inextricably linked to those of their child, and need to be understood in the context of parenting **and** caregiving. A process capable of identifying and addressing parents’ support needs in practice needs to be established.

References

Abell, J., Locke, A., Condor, S., Gibson, S. & Stevenson, C. 2006. Trying similarity, doing difference: the role of interviewer self-disclosure in interview talk with young people. *Qualitative Research,* 6**,** 221-244.

Abernethy, A. P., Capell, W. H., Aziz, N. M., Ritchie, C., Prince-Paul, M., Bennett, R. E. & Kutner, J. S. 2014. Ethical conduct of palliative care research: enhancing communication between investigators and institutional review boards. *J Pain Symptom Manage,* 48**,** 1211-21.

Altınsoy, F. & Aypay, A. 2023. A post-traumatic growth model: psychological hardiness, happiness-increasing strategies, and problem-focused coping. *Current Psychology,* 42**,** 2208-2220.

Aoun, S. M., Stegmann, R., Deleuil, R., Momber, S., Cuddeford, L., Phillips, M. B., Lyon, M. E. & Gill, F. J. 2022. “It Is a Whole Different Life from the Life I Used to Live”: Assessing Parents’ Support Needs in Paediatric Palliative Care. *Children,* 9**,** 322.

Appelbaum, M. G. & Smolowitz, J. L. 2012. Appreciating life: being the father of a child with severe cerebral palsy. *J Neurosci Nurs,* 44**,** 36-42.

Arias-Rojas, M., Carreño-Moreno, S. & Posada-López, C. 2019. Uncertainty in illness in family caregivers of palliative care patients and associated factors. *Revista latino-americana de enfermagem,* 27**,** e3200-e3200.

Arora, S., Goodall, S., Viney, R., Einfeld, S. & The, M. T. 2020. Health-related quality of life amongst primary caregivers of children with intellectual disability. *Journal of Intellectual Disability Research,* 64**,** 103-116.

Avery, M., Wolfe, J. & Decourcey, D. D. 2024. Economic Hardship at the End of Life for Families of Children With Complex Chronic Conditions. *Journal of Pain and Symptom Management,* 67**,** e313-e319.

Bailey-Pearce, O., Stedmon, J., Dallos, R. & Davis, G. 2017. Fathers’ experiences of their child’s life-limiting condition: An attachment narrative perspective. *Clinical Child Psychology and Psychiatry,* 23**,** 381-397.

Bailey-Rodriguez, D. 2021. Qualitatively driven mixed-methods approaches to counselling and psychotherapy research. *Counselling and Psychotherapy Research,* 21**,** 143-153.

Baker, K. & Claridge, A. M. 2023. “I have a Ph.D. in my daughter”: Mother and Child Experiences of Living with Childhood Chronic Illness. *Journal of Child and Family Studies,* 32**,** 2625-2636.

Bally, J. M. G., Burles, M., Smith, N. R., Holtslander, L., Mpofu, C., Hodgson-Viden, H. & Zimmer, M. 2020. Exploring opportunities for holistic family care of parental caregivers of children with life-threatening or life-limiting illnesses. *Qualitative Social Work***,** 1473325020967739.

Bally, J. M. G., Smith, N. R., Holtslander, L., Duncan, V., Hodgson-Viden, H., Mpofu, C. & Zimmer, M. 2018. A Metasynthesis: Uncovering What Is Known About the Experiences of Families With Children Who Have Life-limiting and Life-threatening Illnesses. *J Pediatr Nurs,* 38**,** 88-98.

Barnett-Page, E. & Thomas, J. 2009. Methods for the synthesis of qualitative research: a critical review. *BMC Med Res Methodol,* 9**,** 59.

Barroso, J., Gollop, C. J., Sandelowski, M., Meynell, J., Pearce, P. F. & Collins, L. J. 2003. The challenges of searching for and retrieving qualitative studies. *West J Nurs Res,* 25**,** 153-78.

Bender, J. L., Cyr, A. B., Arbuckle, L. & Ferris, L. E. 2017. Ethics and Privacy Implications of

Using the Internet and Social Media to Recruit Participants for Health Research: A Privacy-by-Design Framework for Online Recruitment. *J Med Internet Res,* 19**,** e104.

Bengtsson, M. 2016. How to plan and perform a qualitative study using content analysis. *NursingPlus Open,* 2**,** 8-14.

Bluebond-Langner, M., Belasco, J. B., Goldman, A. & Belasco, C. 2007. Understanding parents' approaches to care and treatment of children with cancer when standard therapy has failed. *Journal of Clinical Oncology,* 25**,** 2414-2419.

Bluebond-Langner, M., Hargrave, D., Henderson, E., M. & Langner, R. 2017. 'I have to live with the decisions I make': laying a foundation for decision making for children with life-limiting conditions and life-threatening illnesses. *Archives of Disease in Childhood,* 102**,** 468.

Boeije, H. R., Van Wesel, F. & Alisic, E. 2011. Making a difference: towards a method for weighing the evidence in a qualitative synthesis. *J Eval Clin Pract,* 17**,** 657-63.

Booth, A. 2016. Searching for qualitative research for inclusion in systematic reviews: a structured methodological review. *Syst Rev,* 5**,** 74.

Booth, A., Harris, J., Croot, E., Springett, J., Campbell, F. & Wilkins, E. 2013. Towards a methodology for cluster searching to provide conceptual and contextual "richness" for systematic reviews of complex interventions: case study (CLUSTER). *BMC Medical Research Methodology,* 13**,** 118.

Booth, A., Noyes, J., Flemming, K., Gerhardus, A., Wahlster, P., Van Der Wilt, G. J., Mozygemba, K., Refolo, P., Sacchini, D., Tummers, M. & Rehfuess, E. 2018. Structured methodology review identified seven (RETREAT) criteria for selecting qualitative evidence synthesis approaches. *J Clin Epidemiol,* 99**,** 41-52.

Bose, B., Quiñones, F., Moreno, G., Raub, A., Huh, K. & Heymann, J. 2020. Protecting Adults With Caregiving Responsibilities From Workplace Discrimination: Analysis of National Legislation. *Journal of Marriage and Family,* 82**,** 953-964.

Bowlby, J. 1969. *Attachment and Loss*, Basic Books.

Brady, P. W., Giambra, B. K., Sherman, S. N., Clohessy, C., Loechtenfeldt, A. M., Walsh, K. E., Shah, S. S. & Lannon, C. 2020. The Parent Role in Advocating for a Deteriorating Child: A Qualitative Study. *Hospital Pediatrics,* 10**,** 728-742.

Brannan, A. M., Brennan, E. M., Sellmaier, C. & Rosenzweig, J. M. 2018. Employed Parents of Children Receiving Mental Health Services: Caregiver Strain and Work–Life Integration. *Families in Society,* 99**,** 29-44.

Braun, V. & Clarke, V. 2006. Using thematic analysis in psychology. *Qualitative Research in Psychology,* 3**,** 77-101.

Braun, V. & Clarke, V. 2019. Reflecting on reflexive thematic analysis. *Qualitative Research in Sport, Exercise and Health,* 11**,** 589-597.

Braun, V. & Clarke, V. 2020. One size fits all? What counts as quality practice in (reflexive) thematic analysis? *Qualitative Research in Psychology***,** 1-25.

Braun, V. & Clarke, V. 2021a. One size fits all? What counts as quality practice in (reflexive) thematic analysis? *Qualitative Research in Psychology,* 18**,** 328-352.

Braun, V. & Clarke, V. 2021b. To saturate or not to saturate? Questioning data saturation as a useful concept for thematic analysis and sample-size rationales. *Qualitative Research in Sport, Exercise and Health,* 13**,** 201-216.

Braun, V. & Clarke, V. 2024. Supporting best practice in reflexive thematic analysis reporting in Palliative Medicine: A review of published research and introduction to the

Reflexive Thematic Analysis Reporting Guidelines (RTARG). *Palliative Medicine*, 02692163241234800.

Bright, M. A., Franich-Ray, C., Anderson, V., Northam, E., Cochrane, A., Menahem, S. & Jordan, B. 2013. Infant cardiac surgery and the father-infant relationship: feelings of strength, strain, and caution. *Early Hum Dev,* 89**,** 593-9.

Brody, A. C. & Simmons, L. A. 2007. Family resiliency during childhood cancer: the father's perspective. *J Pediatr Oncol Nurs,* 24**,** 152-65.

Bruce, E., Lindh, V. & Sundin, K. 2016. Support for Fathers of Children With Heart Defects. *Clin Nurs Res,* 25**,** 254-72.

Bryman, A. 2007. Barriers to Integrating Quantitative and Qualitative Research. *Journal of Mixed Methods Research,* 1**,** 8-22.

Buchanan, I. 2018. *A Dictionary of Critical Theory,* Online Version, Oxford University Press.

Butler, A., Hall, H. & Copnell, B. 2016. A Guide to Writing a Qualitative Systematic Review Protocol to Enhance Evidence-Based Practice in Nursing and Health Care. *Worldviews Evid Based Nurs,* 13**,** 241-9.

Byrne, D. 2022. A worked example of Braun and Clarke's approach to reflexive thematic analysis. *Quality & Quantity,* 56**,** 1391-1412.

Byun, E., Lerdal, A., Gay, C. L. & Lee, K. A. 2016. How Adult Caregiving Impacts Sleep: a Systematic Review. *Current Sleep Medicine Reports,* 2**,** 191-205.

Campbell, K. A., Orr, E., Durepos, P., Nguyen, L., Li, L., Whitmore, C., Gehrke, P., Graham, L. & Jack, S. M. 2021. Reflexive thematic analysis for applied qualitative health research. *The Qualitative Report,* 26**,** 2011-2028.

Carers UK (2010). Half a million voices: improving support for BAME carers. London: Carers UK.

Carmassi, C., Corsi, M., Bertelloni, C. A., Carpita, B., Gesi, C., Pedrinelli, V., Massimetti, G., Peroni, D. G., Bonuccelli, A., Orsini, A. & Dell'osso, L. 2018. Mothers and fathers of children with epilepsy: gender differences in post-traumatic stress symptoms and correlations with mood spectrum symptoms. *Neuropsychiatric Disease and Treatment,* 14**,** 1371-1379.

Carroll, C., Booth, A. & Lloyd-Jones, M. 2012. Should we exclude inadequately reported studies from qualitative systematic reviews? An evaluation of sensitivity analyses in two case study reviews. *Qual Health Res,* 22**,** 1425-34.

Casp 2018. Critical Appraisal Skills Programme (2019). CASP Qualitative Checklist. [online] Available at: https://casp-uk.net/wp-content/uploads/2018/01/CASP-Qualitative-Checklist-2018.pdf.

Chamberlain, H. J. 2007. *Fathers' resilience in the face of their child's leukemia : a qualitative study.* Alliant International University.

Chesler, M. A. & Parry, C. 2001. Gender roles and/or styles in crisis: An integrative analysis of the experiences of fathers of children with cancer. *Qualitative Health Research,* 11**,** 363-384.

Choi, S. & Seo, J. 2019. Analysis of caregiver burden in palliative care: An integrated review. *Nursing Forum,* 54**,** 280-290.

Choonara, J., Murgia, A. & Carmo, R. M. 2022. *Faces of Precarity*

Critical Perspectives on Work, Subjectivities and Struggles, Bristol University Press.

Chung, H. & Van Der Lippe, T. 2020. Flexible Working, Work–Life Balance, and Gender Equality: Introduction. *Social Indicators Research,* 151**,** 365-381.

Clark, S. M. & Miles, M. S. 1999. Conflicting responses: The experiences of fathers of infants diagnosed with severe congenital heart disease. *Journal of the Society of Pediatric Nurses,* 4**,** 7-14.

Clarke, J. N. 2005. Fathers' Home Health Care Work When a Child Has Cancer: I'm Her Dad; I Have to Do It. *Men and Masculinities,* 7**,** 385-404.

Cluley, S. M. 2015. *Fathers of children with cancer : a narrative inquiry : a thesis presented in partial fultilment of the requirements for the degree of Doctor of Clinical Psychology at Massey University, Albany, New Zealand.* Doctor of Clinical Psychology (D. Clin. Psych.) Doctoral, Massey University.

Collins, A., Hennessy-Anderson, N., Hosking, S., Hynson, J., Remedios, C. & Thomas, K. 2016. Lived experiences of parents caring for a child with a life-limiting condition in Australia: A qualitative study. *Palliat Med,* 30**,** 950-959.

Collins, A. B., J.; Remedios, C.; & Thomas, K. 2020. Describing the psychosocial profile and unmet support needs of parents caring for a child with a life-limiting condition: A cross-sectional study of caregiver-reported outcomes. *Palliative Medicine,* 34**,** 358–366.

Conijn, T., De Roos, C., Vreugdenhil, H. J. I., Van Dijk-Lokkart, E. M., Wijburg, F. A. & Haverman, L. 2022. Effectiveness of time-limited eye movement desensitization reprocessing therapy for parents of children with a rare life-limiting illness: a randomized clinical trial. *Orphanet Journal of Rare Diseases,* 17**,** 328.

Connell, J., O'cathain, A. & Brazier, J. 2014. Measuring quality of life in mental health: Are we asking the right questions? *Social Science & Medicine,* 120**,** 12-20.

Cooke, A., Smith, D. & Booth, A. 2012. Beyond PICO: the SPIDER tool for qualitative evidence synthesis. *Qual Health Res,* 22**,** 1435-43.

Cooper, B., Kinsella, G. J. & Picton, C. 2006. Development and initial validation of a family appraisal of caregiving questionnaire for palliative care. *Psycho-Oncology,* 15**,** 613-622.

Corlett, J. & Twycross, A. 2006. Negotiation of parental roles within family-centred care: a review of the research. *Journal of Clinical Nursing,* 15**,** 1308-1316.

Courtney, E., Kiernan, G., Guerin, S., Ryan, K. & Mcquillan, R. 2018. Mothers' perspectives of the experience and impact of caring for their child with a life-limiting neurodevelopmental disability. *Child: Care, Health and Development,* 44**,** 704-710.

Coventry, P., Taylor, J., Overend, K., Chew-Graham, C., Knowles, S., Gilbody, S., Lewis, H. & Littlewood, L. 2016. Understanding how collaborative care is effectively implemented for treating common mental health problems: a qualitative meta-synthesis of provider and patient perspectives. PROSPERO 2016 CRD42016037561. Available from: https://www.crd.york.ac.uk/prospero/display_record.php?ID=CRD42016037561.

Creary, S. J. & Gordon, J. R. 2016. Role Conflict, Role Overload, and Role Strain. *Encyclopedia of Family Studies.*

Creswell, J. W. & Clark, V. L. P. 2007. *Designing and conducting mixed methods research,* Thousand Oaks, CA, US, Sage Publications, Inc.

Creswell, J. W. & Creswell, J. D. 2017. *Research design: Qualitative, quantitative, and mixed methods approaches*, Sage publications.

Creswell, J. W. & Plano Clark, V. L. 2011. *Designing and conducting mixed methods research* Los Angeles, Calif, SAGE Publications.

Darko, E. M., Kleib, M. & Olson, J. 2022. Social Media Use for Research Participant Recruitment: Integrative Literature Review. *J Med Internet Res,* 24**,** e38015.

Davies, B., Baird, J. & Gudmundsdottir, M. 2013. Moving family-centered care forward: Bereaved fathers' perspectives. *J Hosp Palliat Nurs,* 15.

Davies, B., Gudmundsdottir, M., Worden, B., Orloff, S., Sumner, L. & Brenner, P. 2004. "Living in the dragon's shadow" fathers' experiences of a child's life-limiting illness. *Death Stud,* 28**,** 111-35.

Davison, K. K., Charles, J. N., Khandpur, N. & Nelson, T. J. 2017. Fathers' Perceived Reasons for Their Underrepresentation in Child Health Research and Strategies to Increase Their Involvement. *Matern Child Health J,* 21**,** 267-274.

De Groot, J. M., Fehon, D. C., Calman, L., Miller, D. S. & Feldstain, A. 2023. Trauma-informed palliative care is needed: A call for implementation and research. *Palliative Medicine,* 37**,** 1470-1473.

Deave, T. & Johnson, D. 2008. The transition to parenthood: what does it mean for fathers? *Journal of Advanced Nursing,* 63**,** 626-633.

Demerouti, E., Bakker, A. B., Nachreiner, F. & Schaufeli, W. B. 2001. The job demands-resources model of burnout. *Journal of Applied psychology,* 86**,** 499.

Denscombe, M. 2008. Communities of Practice: A Research Paradigm for the Mixed Methods Approach. *Journal of Mixed Methods Research,* 2**,** 270-283.

Diniz, E., Brandão, T., Monteiro, L. & Veríssimo, M. 2021. Father Involvement During Early Childhood: A Systematic Review of the Literature. *Journal of Family Theory & Review,* 13**,** 77-99.

Dures, E., Rumsey, N., Morris, M. & Gleeson, K. 2010. Mixed Methods in Health Psychology: Theoretical and Practical Considerations of the Third Paradigm. *Journal of Health Psychology,* 16**,** 332-341.

Eggenberger, L., Ehlert, U. & Walther, A. 2023. New directions in male-tailored psychotherapy for depression. *Frontiers in Psychology,* 14.

Einstein, D. A. 2014. Extension of the Transdiagnostic Model to Focus on Intolerance of Uncertainty: A Review of the Literature and Implications for Treatment. *Clinical Psychology: Science and Practice,* 21**,** 280-300.

Ekberg, S. B., Nk; Herbert, a; Danby, S; Yates, P; 2018. Healthcare Users' Experiences of Communicating with Healthcare Professionals About Children Who Have Life-Limiting Conditions: A Qualitative Systematic Review. *Journal of Palliative Medicine,* 21**,** 1518-1528.

Euroqol. https://euroqol.org/eq-5d-instruments/eq-5d-5l-about/valuation-standard-value-sets/

Fàbregues, S., Escalante-Barrios, E. L., Molina-Azorin, J. F., Hong, Q. N. & Verd, J. M. 2021. Taking a critical stance towards mixed methods research: A cross-disciplinary qualitative secondary analysis of researchers' views. *PLoS One,* 16**,** e0252014.

Fàbregues, S., Hong, Q. N., Escalante-Barrios, E. L., Guetterman, T. C., Meneses, J. & Fetters, M. D. 2020. A Methodological Review of Mixed Methods Research in Palliative and End-of-Life Care (2014-2019). *Int J Environ Res Public Health,* 17.

Fakis, A., Hilliam, R., Stoneley, H. & Townend, M. 2014. Quantitative Analysis of Qualitative Information From Interviews:A Systematic Literature Review. *Journal of Mixed Methods Research,* 8**,** 139-161.

Fetters, M. D., Curry, L. A. & Creswell, J. W. 2013. Achieving integration in mixed methods designs-principles and practices. *Health Serv Res,* 48**,** 2134-56.

Feudtner, C., Nye, R. T., Boyden, J. Y., Schwartz, K. E., Korn, E. R., Dewitt, A. G., Waldman, A. T., Schwartz, L. A., Shen, Y. A., Manocchia, M., Xiao, R., Lord, B. T. & Hill, D. L. 2021. Association Between Children With Life-Threatening Conditions and Their Parents' and Siblings' Mental and Physical Health. *JAMA Network Open,* 4**,** e2137250-e2137250.

Fisher, V., Atkin, K., Ewing, G., Grande, G. & Fraser, L. K. 2023. Assessing the suitability of the Carer Support Needs Assessment Tool (CSNAT-Paediatric) for use with parents of children with a life-limiting condition: A qualitative secondary analysis. *Palliative Medicine,* 38**,** 100-109.

Flanagin, A., Frey, T., Christiansen, S. L. & Committee, A. M. O. S. 2021. Updated Guidance on the Reporting of Race and Ethnicity in Medical and Science Journals. *JAMA,* 326**,** 621-627.

Flemming, K., Booth, A., Garside, R., Tuncalp, O. & Noyes, J. 2019. Qualitative evidence synthesis for complex interventions and guideline development: clarification of the purpose, designs and relevant methods. *BMJ Glob Health,* 4**,** e000882.

Flemming, K. & Briggs, M. 2007. Electronic searching to locate qualitative research: evaluation of three strategies. *J Adv Nurs,* 57**,** 95-100.

Flemming, K., Turner, V., Bolsher, S., Hulme, B., Mchugh, E. & Watt, I. 2020. The experiences of, and need for, palliative care for people with motor neurone disease and their informal caregivers: A qualitative systematic review. *Palliative Medicine,* 34**,** 708-730.

Flick, U. 2017. *The SAGE Handbook of Qualitative Data Collection*, SAGE Publications.

Fortune, A., Perkins, E., Paize, F., Palanisami, B. & Gladstone, M. 2024. Managing mothers' and fathers' uncertainty during their journey through early neurodevelopmental follow-up for their high-risk infants—A qualitative account. *Child: Care, Health and Development,* 50**,** e13168.

Fraser, L. K., Gibson Smith, D., Jarvis, S., Norman, P. & Parslow, R. 2020a. 'Make Every Child Count' Estimating current and future prevalence of children and young people with life-limiting conditions in the United Kingdom.

Fraser, L. K., Gibson-Smith, D., Jarvis, S., Norman, P. & Parslow, R. C. 2020b. Estimating the current and future prevalence of life-limiting conditions in children in England. *Palliative Medicine***,** 0269216320975308.

Fraser, L. K., Jarvis, S., Moran, N., Aldridge, J., Parslow, R. & Beresford, B. 2015. Children in Scotland requiring Palliative Care: identifying numbers and needs (The ChiSP Study).

Fraser, L. K., Murtagh, F. E. M., Aldridge, J., Sheldon, T., Gilbody, S. & Hewitt, C. 2021. Health of mothers of children with a life-limiting condition: a comparative cohort study. *Archives of Disease in Childhood***,** archdischild-2020-320655.

Gallagher, S., Phillips, A. C. & Carroll, D. 2010. Parental stress is associated with poor sleep quality in parents caring for children with developmental disabilities. *Journal of Pediatric Psychology,* 35**,** 728-737.

Gelinas, L., Pierce, R., Winkler, S., Cohen, I. G., Lynch, H. F. & Bierer, B. E. 2017. Using Social Media as a Research Recruitment Tool: Ethical Issues and Recommendations. *The American Journal of Bioethics,* 17**,** 3-14.

Georgas, J. 2004. Family and Culture. *In:* SPIELBERGER, C. D. (ed.) *Encyclopedia of Applied Psychology.* New York: Elsevier.

Gérain, P. & Zech, E. 2019. Informal Caregiver Burnout? Development of a Theoretical Framework to Understand the Impact of Caregiving. *Frontiers in Psychology,* 10.

Gérain, P. & Zech, E. 2022. Are caregiving appraisal and relationship quality key mediators in informal caregiving burnout? A structural equation modelling study in Belgium and France. *Health & Social Care in the Community,* 30**,** e2433-e2444.

Gibson-Smith, D., Jarvis, S. W. & Fraser, L. K. 2021. Place of death of children and young adults with a life-limiting condition in England: a retrospective cohort study. *Archives of Disease in Childhood,* 106**,** 780.

Gill, F. J., Hashem, Z., Stegmann, R. & Aoun, S. M. 2020. The support needs of parent caregivers of children with a life-limiting illness and approaches used to meet their needs: A scoping review. *Palliative Medicine***,** 0269216320967593.

Government, S. 2018. Scottish Household Survey.

Gower, C., Higgins, A., Doherty, N. & Mccormack, D. 2016. Understanding the experiences of fathers of children with congenital heart disease: An interpretative phenomenological analysis. *Journal of Health Psychology,* 22**,** 1447-1457.

Greenwood, N., Mckevitt, C. & Milne, A. 2018. Time to rebalance and reconsider: are we pathologising informal, family carers? *Journal of the Royal Society of Medicine,* 111**,** 253-254.

Hain, R., Devins, M., Hastings, R. & Noyes, J. 2013. Paediatric palliative care: development and pilot study of a 'Directory' of life-limiting conditions. *BMC Palliat Care,* 12**,** 43.

Hajek, A., Kretzler, B. & König, H.-H. 2021. Informal Caregiving, Loneliness and Social Isolation: A Systematic Review. *International Journal of Environmental Research and Public Health* [Online], 18.

Halinski, M., Duxbury, L. & Stevenson, M. 2020. Employed Caregivers' Response to Family-Role Overload: the Role of Control-at-Home and Caregiver Type. *Journal of Business and Psychology,* 35**,** 99-115.

Hanish, A. E., Lin-Dyken, D. C. & Han, J. C. 2017. PROMIS Sleep Disturbance and Sleep-Related Impairment in Adolescents: Examining Psychometrics Using Self-Report and Actigraphy. *Nursing research,* 66**,** 246-251.

Harris, J. L., Booth, A., Cargo, M., Hannes, K., Harden, A., Flemming, K., Garside, R., Pantoja, T., Thomas, J. & Noyes, J. 2018. Cochrane Qualitative and Implementation Methods Group guidance series-paper 2: methods for question formulation, searching, and protocol development for qualitative evidence synthesis. *J Clin Epidemiol,* 97**,** 39-48.

Hayes, C. C. & Savage, E. 2008. Fathers' perspectives on the emotional impact of managing the care of their children with cystic fibrosis. *J Pediatr Nurs,* 23**,** 250-6.

Hearn, G., Clarkson, G. & Day, M. 2020. The Role of the NICU in Father Involvement, Beliefs, and Confidence: A Follow-up Qualitative Study. *Advances in Neonatal Care,* 20.

Hecht, L. M. 2001. Role Conflict and Role Overload: Different Concepts, Different Consequences. *Sociological Inquiry,* 71**,** 111-121.

Herdman, M., Gudex, C., Lloyd, A., Janssen, M., Kind, P., Parkin, D., Bonsel, G. & Badia, X. 2011. Development and preliminary testing of the new five-level version of EQ-5D (EQ-5D-5L). *Qual Life Res,* 20**,** 1727-36.

Hill, K. & Coyne, I. 2012. Palliative care nursing for children in the UK and Ireland. *British Journal of Nursing,* 21**,** 276-281.

Hill, K., Higgins, A., Dempster, M. & Mccarthy, A. 2009. Fathers' views and understanding of their roles in families with a child with acute lymphoblastic leukaemia: an interpretative phenomenological analysis. *J Health Psychol,* 14**,** 1268-80.

Hillman, A., Jones, I. R., Quinn, C., Pentecost, C., Stapley, S., Charlwood, C. & Clare, L. 2023. The precariousness of living with, and caring for people with, dementia: Insights from the IDEAL programme. *Social Science & Medicine,* 331**,** 116098.

Hoell, J. I., Weber, H., Warfsmann, J., Trocan, L., Gagnon, G., Danneberg, M., Balzer, S., Keller, T., Janßen, G. & Kuhlen, M. 2019. Facing the large variety of life-limiting conditions in children. *European Journal of Pediatrics,* 178**,** 1893-1902.

Hovey, J. K. 2005. Fathers Parenting Chronically Ill Children: Concerns and Coping Strategies. *Issues in Comprehensive Pediatric Nursing,* 28**,** 83-95.

Hurley, F., Kiernan, G. & Price, J. 2021. 'Starting Out in Haziness': Parental Experiences Surrounding the Diagnosis of their Child's Non-Malignant Life-Limiting Condition in Ireland. *Journal of Pediatric Nursing,* 59**,** 25-31.

Hwang, B., Granger, D. A., Brecht, M.-L. & Doering, L. V. 2022. Cognitive behavioral therapy versus general health education for family caregivers of individuals with heart failure: a pilot randomized controlled trial. *BMC Geriatrics,* 22**,** 281.

Jaaniste, T., Cuganesan, A., Chin, W. L. A., Tan, S. C., Coombs, S., Heaton, M., Cowan, S., Aouad, P., Potter, D., Smith, P. L. & Trethewie, S. 2021. Living with a child who has a life-limiting condition: The functioning of well-siblings and parents. *Child Care Health Dev*.

Jarvis, S. W., Roberts, D., Flemming, K., Richardson, G. & Fraser, L. K. 2021. Transition of children with life-limiting conditions to adult care and healthcare use: a systematic review. *Pediatric Research*.

Jingchu, H., Biao, F., Yonghui, Z., Wenqing, W., Jiawei, X. & Xifu, Z. 2017. Gender Differences in PTSD: Susceptibility and Resilience. *In:* AIDA, A. (ed.) *Gender Differences in Different Contexts.* Rijeka: IntechOpen.

Johansson, T. 2023. Theorising fatherhood: challenges and suggestions. *Families, Relationships and Societies,* 12**,** 49-59.

Johnson, R. & Christensen, L. 2014. *Educational Research Quantitative, Qualitative, and Mixed Approaches Fifth Edition*.

Jones, B. L., Pelletier, W., Decker, C., Barczyk, A. & Dungan, S. S. 2010. Fathers of children with cancer: a descriptive synthesis of the literature. *Soc Work Health Care,* 49**,** 458-93.

Jones, J. B. & Neil-Urban, S. 2003. Father to father: focus groups of fathers of children with cancer. *Soc Work Health Care,* 37**,** 41-61.

Kaufman, G. 2018. Barriers to equality: why British fathers do not use parental leave. *Community, Work & Family,* 21**,** 310-325.

Kaye, E. C., Gushue, C. A., Demarsh, S., Jerkins, J., Li, C., Lu, Z., Snaman, J. M., Blazin, L., Johnson, L.-M., Levine, D. R., Morrison, R. R. & Baker, J. N. 2019. Impact of Race and Ethnicity on End-of-Life Experiences for Children With Cancer. *American Journal of Hospice and Palliative Medicine®,* 36**,** 767-774.

Kazak, A. E., Kassam-Adams, N., Schneider, S., Zelikovsky, N., Alderfer, M. A. & Rourke, M. 2005. An Integrative Model of Pediatric Medical Traumatic Stress. *Journal of Pediatric Psychology,* 31**,** 343-355.

Kim, H. & Rose, K. 2011. Sleep disturbances in family caregivers: an overview of the state of the science. *Arch Psychiatr Nurs,* 25**,** 456-68.

Kim, T. H. M., Delahunty-Pike, A. & Campbell-Yeo, M. 2020. Effect of Fathers' Presence and Involvement in Newborn Care in the NICU on Mothers' Symptoms of Postpartum Depression. *Journal of Obstetric, Gynecologic & Neonatal Nursing,* 49**,** 452-463.

King, G., King, S., Rosenbaum, P. & Goffin, R. 1999. Family-centered caregiving and well-being of parents of children with disabilities:linking process with outcome. *Journal of Pediatric Psychology,* 24**,** 41-53.

Klapproth, C. P., Sidey-Gibbons, C. J., Valderas, J. M., Rose, M. & Fischer, F. 2022. Comparison of the PROMIS Preference Score (PROPr) and EQ-5D-5L Index Value in General Population Samples in the United Kingdom, France, and Germany. *Value in Health,* 25**,** 824-834.

Knights, D. 2019. Gender still at work: Interrogating identity in discourses and practices of masculinity. *Gender, Work & Organization,* 26**,** 18-30.

Koch, A., Kozhumam, A. S., Seeler, E., Docherty, S. L. & Brandon, D. 2021. Multiple roles of parental caregivers of children with complex life-threatening conditions: A qualitative descriptive analysis. *Journal of Pediatric Nursing,* 61**,** 67-74.

Koch, K. D. & Jones, B. L. 2018. Supporting Parent Caregivers of Children with Life-Limiting Illness. *Children (Basel),* 5.

Kokurcan, A., Özpolat, A. G. Y. & Göğüş, A. K. 2015. Burnout in caregivers of patients with schizophrenia. *Turkish journal of medical sciences,* 45**,** 678-685.

Lain, D., Airey, L., Loretto, W. & Vickerstaff, S. 2019. Understanding older worker precarity: the intersecting domains of jobs, households and the welfare state. *Ageing and Society,* 39**,** 2219-2241.

Lamb, M. 1975. Fathers: Forgotten Contributors to Child Development. *Human development,* 18**,** 245-66.

Law, S., Ormel, I., Babinski, S., Plett, D., Dionne, E., Schwartz, H. & Rozmovits, L. 2021. Dread and solace: Talking about perinatal mental health. *Int J Ment Health Nurs,* 30 Suppl 1**,** 1376-1385.

Laws, T. 2018. Supporting fathers who want to be involved in providing healthcare for their child. *Nurs Child Young People,* 30**,** 18-26.

Lawton, M. P., Moss, M., Kleban, M. H., Glicksman, A. & Rovine, M. 1991. A two-factor model of caregiving appraisal and psychological well-being. *J Gerontol,* 46**,** P181-9.

Lazarus, R., A. & Folkman, S. 1984. *Stress, appraisal, and coping,* New York, Springer.

Lazarus, R. S. 1966. *Psychological Stress and the Coping Process,* New York, McGraw-Hill.

Lazarus, R. S. 1996. *The role of coping in the emotions and how coping changes over the life course*, Elsevier.

Lee, J., Baik, S., Becker, T. D. & Cheon, J. H. 2021. Themes describing social isolation in family caregivers of people living with dementia: A scoping review. *Dementia,* 21**,** 701-721.

Lee, R. T. & Ashforth, B. E. 1990. On the meaning of Maslach's three dimensions of burnout. *J Appl Psychol,* 75**,** 743-7.

Leech, N. L. & Onwuegbuzie, A. J. 2009. A typology of mixed methods research designs. *Quality & Quantity: International Journal of Methodology,* 43**,** 265-275.

Leung, L. 2015. Validity, reliability, and generalizability in qualitative research. *J Family Med Prim Care,* 4**,** 324-7.

Lewin, S., Glenton, C., Munthe-Kaas, H., Carlsen, B., Colvin, C. J., Gulmezoglu, M., Noyes, J., Booth, A., Garside, R. & Rashidian, A. 2015. Using qualitative evidence in decision making for health and social interventions: an approach to assess confidence in findings from qualitative evidence syntheses (GRADE-CERQual). *PLoS Med,* 12**,** e1001895.

Li, Q., Zhang, H., Zhang, M., Li, T., Ma, W., An, C., Chen, Y., Liu, S., Kuang, W., Yu, X. & Wang, H. 2020. Prevalence and Risk Factors of Anxiety, Depression, and Sleep Problems Among Caregivers of People Living With Neurocognitive Disorders During the COVID-19 Pandemic. *Front Psychiatry,* 11**,** 590343.

Lian, B. X., Amin, Z., Sensaki, S. & Aishworiya, R. 2020. An active pursuit of reassurance—coping strategies of fathers with infants in the Neonatal Intensive Care Unit. *Journal of Perinatology*.

Lindahl Norberg, A., Pöder, U., Ljungman, G. & Von Essen, L. 2012. Objective and subjective factors as predictors of post-traumatic stress symptoms in parents of children with cancer--a longitudinal study. *PLoS One,* 7**,** e36218.

Liu, Y., Sundquist, J., Sundquist, K., Zheng, D. & Ji, J. 2023. Mental health outcomes in parents of children with a cancer diagnosis in Sweden: a nationwide cohort study. *eClinicalMedicine,* 55**,** 101734.

Llosa, JA., Agulló-Tomás, E., Menéndez-Espina, S., Rivero-Díaz, ML. 2024 Perceived precarious life: a SEM model for re-dimensioning of precarious work and its impact on mental health. *Front Public Health*. 5;11:1254843. doi: 10.3389/fpubh.2023.1254843

Long, H. A., French, D. P. & Brooks, J. M. 2020. Optimising the value of the critical appraisal skills programme (CASP) tool for quality appraisal in qualitative evidence synthesis. *Research Methods in Medicine & Health Sciences,* 1**,** 31-42.

Lucca, S. A. & Petean, E. B. 2016. Fatherhood: experiences of fathers of boys diagnosed with Duchenne Muscular Dystrophy. *Cien Saude Colet,* 21**,** 3081-3089.

Mackay, G. 2016. Is there a need to Differentiate between Qualitative and Quantitative Searching Strategies for Literature Reviewing? *Qualitative Social Work: Research and Practice,* 6**,** 231-241.

Major, D. A. 2003. Utilizing role theory to help employed parents cope with children's chronic illness. *Health Education Research,* 18**,** 45-57.

Manzari, Z. S., Rafiei, H., Ghaderi, M. S., Abedi, F. & Mafi, M. H. 2023. Relationship between Resilience and Caregiver Burden Among Home Caregivers of COVID-19 Patients. *Home Healthc Now,* 41**,** 42-48.

Martin, L. A., Neighbors, H. W. & Griffith, D. M. 2013. The Experience of Symptoms of Depression in Men vs Women: Analysis of the National Comorbidity Survey Replication. *JAMA Psychiatry,* 70**,** 1100-1106.

Mason, J. 2006. Mixing methods in a qualitatively driven way. *Qualitative Research,* 6**,** 9-25.

Maxwell, J. A. 2021. Why qualitative methods are necessary for generalization. *Qualitative Psychology,* 8**,** 111-118.

Mays, N., Pope, C. & Popay, J. 2005. Systematically reviewing qualitative and quantitative evidence to inform management and policy-making in the health field. *Journal of Health Services Research & Policy,* 10**,** 6-20.

Mccann, D., Bull, R. & Winzenberg, T. 2014. Sleep Deprivation in Parents Caring for Children With Complex Needs at Home: A Mixed Methods Systematic Review. *Journal of family nursing,* 21.

Mcdonald, T. P., Gregoire, T. K., Poertner, J. & Early, T. J. 1997. Building a model of family caregiving for children with emotional disorders. *Journal of Emotional and Behavioral Disorders,* 5**,** 138-148.

Mcgeehin Heilferty, C. 2018. The Search for Balance: Prolonged Uncertainty in Parent Blogs of Childhood Cancer. *Journal of Family Nursing,* 24**,** 250-270.

Mckenzie, J., Brennan, S., Ryan, R., Thomson, H., Johnston, R. & Thomas, J. 2019. Chapter 3: Defining the criteria for including studies and how they will be grouped for the synthesis. *In:* HIGGINS, J., THOMAS, J., CHANDLER, J., CUMPSTON, M., LI, T., PAGE, M. & WELCH, V. (eds.) *Cochrane Handbook for Systematic Reviews of Interventions version 6.0 (updated July 2019).* Available from www.training.cochrane.org/handbook.: Cochrane.

Mcmullin, C. 2021. Transcription and Qualitative Methods: Implications for Third Sector Research. *VOLUNTAS: International Journal of Voluntary and Nonprofit Organizations*.

Mezgebu, E., Berhan, E. & Deribe, L. 2020. Predictors of Resilience Among Parents of Children with Cancer: Cross-Sectional Study. *Cancer Management and Research,* 12**,** 11611-11621.

Mitchell, S., Bennett, K., Morris, A., Slowther, A.-M., Coad, J. & Dale, J. 2019a. Achieving beneficial outcomes for children with life-limiting and life-threatening conditions receiving palliative care and their families: A realist review. *Palliative Medicine,* 34**,** 387-402.

Mitchell, S., Morris, A., Bennett, K., Sajid, L. & Dale, J. 2017. Specialist paediatric palliative care services: what are the benefits? *Arch Dis Child,* 102**,** 923-929.

Mitchell, S., Slowther, A.-M., Coad, J. & Dale, J. 2021. Experiences of healthcare, including palliative care, of children with life-limiting and life-threatening conditions and their families: a longitudinal qualitative investigation. *Archives of Disease in Childhood,* 106**,** 570.

Mitchell, S., Spry, J. L., Hill, E., Coad, J., Dale, J. & Plunkett, A. 2019b. Parental experiences of end of life care decision-making for children with life-limiting conditions in the paediatric intensive care unit: a qualitative interview study. *BMJ Open,* 9**,** e028548.

Mitchell, S. M. 2021. True Resilience: A Look Inside COVID's Effect on Children with Medical Complexity and Their Families. *Curr Pediatr Rep,* 9**,** 171-177.

Mitterer, S., Zimmermann, K., Bergsträsser, E., Simon, M., Gerber, A.-K. & Fink, G. 2021. Measuring Financial Burden in Families of Children Living With Life-Limiting Conditions: A Scoping Review of Cost Indicators and Outcome Measures. *Value in Health,* 24**,** 1377-1389.

Mojica, N. Men As Caregivers: Latino Fathering of Children with Cancer. 2016.

Mu, P.-F., Ma, F.-C., Hwang, B. & Chao, Y.-M. 2002. Families of Children With Cancer: The Impact on Anxiety Experienced by Fathers. *Cancer Nursing,* 25.

Mullen, J., Kelley, E. & Kelloway, E. K. 2008. CHAPTER 11 - Health and Well-Being Outcomes of the Work-Family Interface. *In:* KORABIK, K., LERO, D. S. & WHITEHEAD, D. L. (eds.) *Handbook of Work-Family Integration.* San Diego: Academic Press.

Najafi, K., Shirinabadi Farahani, A., Rassouli, M., Alavi Majd, H. & Karami, M. 2022. Emotional upheaval, the essence of anticipatory grief in mothers of children with life threatening illnesses: a qualitative study. *BMC Psychology,* 10**,** 196.

Neil-Urban, S. & Jones, J. B. 2002. Father-to-Father Support: Fathers of Children with Cancer Share Their Experience. *Journal of Pediatric Oncology Nursing,* 19**,** 97-103.

Neimann Rasmussen, L. & Montgomery, P. 2018. The prevalence of and factors associated with inclusion of non-English language studies in Campbell systematic reviews: a survey and meta-epidemiological study. *Syst Rev,* 7**,** 129.

Newton, B., Tamkin, P., Gloster, R., Cox, A., Everett, C. & Cotton, J. 2018. Rapid evidence assessment: parents' decisions about returning to work and child caring responsibilities. *Research review. London: Institute for Employment Studies*.

Ni, Z. H., Lv, H. T., Wu, J. H. & Wang, F. 2023. Post-traumatic growth in caregivers of children hospitalized in the PICU due to traffic accident: a qualitative study. *BMC Nursing,* 22**,** 48.

Nice 2016. National Institute for Health and Care Excellence; End of life care for infants, children and young people with life-limiting conditions: planning and management.

Nicholas, D., Beaune, L., Belletrutti, M., Blumberg, J., Ing, S., Rapoport, A. & Barrera, M. 2020. Engaging Fathers in Pediatric Palliative Care Research. *Journal of Social Work in End-of-Life & Palliative Care,* 16**,** 42-56.

Nicholas, D. B., Beaune, L., Barrera, M., Blumberg, J. & Belletrutti, M. 2016. Examining the Experiences of Fathers of Children with a Life-Limiting Illness. *J Soc Work End Life Palliat Care,* 12**,** 126-44.

Nicholas, D. B., Gearing, R. E., Mcneill, T., Fung, K., Lucchetta, S. & Selkirk, E. K. 2009. Experiences and resistance strategies utilized by fathers of children with cancer. *Soc Work Health Care,* 48**,** 260-75.

Noblit, G. & Hare, R. 1988. *Meta-ethnography: synthesizing qualitative studies*, Newbury Park: Sage.

Novianti, R., Suarman & Islami, N. 2023. Parenting in Cultural Perspective

A Systematic Review of Paternal Role Across Cultures. *Journal of Ethnic and Cultural Studies,* 10**,** 22-44.

Nowell, L. S., Norris, J. M., White, D. E. & Moules, N. J. 2017. Thematic Analysis: Striving to Meet the Trustworthiness Criteria. *International Journal of Qualitative Methods,* 16**,** 1609406917733847.

Noyes, J., Booth, A., Cargo, M., Flemming, K., Harden, A., Harris, J., Garside, R., Hannes, K., Pantoja, T. & Thomas, J. 2019. Chapter 21: Qualitative evidence. *In:* HIGGINS, J., THOMAS, J., CHANDLER, J., CUMPSTON, M., LI, T., PAGE, M. & WELCH, V. (eds.) *Cochrane Handbook for Systematic Reviews of Interventions version 6.0.* Cochrane, 2019.

Noyes, J. & Popay, J. 2007. Directly observed therapy and tuberculosis: how can a systematic review of qualitative research contribute to improving services? A qualitative meta-synthesis. *J Adv Nurs,* 57**,** 227-43.

Oakley, S., Dunbar, H. & De Vries, K. 2021. Parent-led strategies supporting personal well-being when caring for a child with a life-limiting condition: A scoping review. *Journal of Child Health Care***,** 13674935211026122.

Ogg, M. G. 1997. The Effects of Pediatric Cancer on Fathers During the Diagnostic and Initial Treatment Phases.

Ogourtsova, T., O'donnell, M. E., Chung, D., Gavin, F., Bogossian, A. & Majnemer, A. 2021. Fathers Matter: Enhancing Healthcare Experiences Among Fathers of Children With Developmental Disabilities. *Front Rehabil Sci,* 2**,** 709262.

Olff, M. 2017. Sex and gender differences in post-traumatic stress disorder: an update. *Eur J Psychotraumatol.,* 8**,** 1351204. doi: 10.1080/20008198.2017.1351204. eCollection 2017.

Pascoe Leahy, C. 2021. The afterlife of interviews: explicit ethics and subtle ethics in sensitive or distressing qualitative research. *Qualitative Research,* 22**,** 777-794.

Pearlin, L. I., Menaghan, E. G., Lieberman, M. A. & Mullan, J. T. 1981. The stress process. *Journal of Health and Social behavior***,** 337-356.

Pearlin, L. I., Mullan, J. T., Semple, S. J. & Skaff, M. M. 1990. Caregiving and the stress process: an overview of concepts and their measures. *Gerontologist,* 30**,** 583-94.

Pelentsov, L. J., Laws, T. A. & Esterman, A. J. 2015. The supportive care needs of parents caring for a child with a rare disease: A scoping review. *Disability and Health Journal,* 8**,** 475-491.

Pentaris, P., Papadatou, D., Jones, A., Hosang, GM. 2018 Palliative care professional's perceptions of barriers and challenges to accessing children's hospice and palliative care services in South East London: A preliminary study. *Death Stud*. 42(10), 649-657.

Perez, M. N., Traino, K. A., Bakula, D. M., Sharkey, C. M., Espeleta, H. C., Delozier, A. M., Mayes, S., Mcnall, R., Chaney, J. M. & Mullins, L. L. 2020. Barriers to care in pediatric cancer: The role of illness uncertainty in relation to parent psychological distress. *Psycho-Oncology,* 29**,** 304-310.

Petts, R. 2022. *Father involvement and gender equality in the United States: Contemporary norms and barriers*, Routledge.

Pfitzner, N., Humphreys, C. & Hegarty, K. 2018. Engaging Men as Fathers: How Gender Influences Men’s Involvement in Australian Family Health Services. *Journal of Family Issues,* 39**,** 3956-3985.

Pinquart, M. 2018. Featured Article: Depressive Symptoms in Parents of Children With Chronic Health Conditions: A Meta-Analysis. *Journal of Pediatric Psychology,* 44**,** 139-149.

Plano Clark, V. I., N. 2016. *Why use mixed methods research?: identifying rationales for mixing methods. In Mixed methods research: A guide to the field* SAGE Publications, Inc.

Poley, M. J., Brouwer, W. B., Van Exel, N. J. & Tibboel, D. 2012. Assessing health-related quality-of-life changes in informal caregivers: an evaluation in parents of children with major congenital anomalies. *Qual Life Res,* 21**,** 849-61.

Polita, N. B., Alvarenga, W. D. A., Leite, A. C. a. B., Araújo, J. S., Santos, L. B. P. a. D., Zago, M. M. F., Montigny, F. D. & Nascimento, L. C. 2018. Care provided by the father to the child with cancer under the influence of masculinities: qualitative meta-synthesis. *Revista Brasileira de Enfermagem,* 71**,** 185-194.

Portacolone, E., Rubinstein, R. L., Covinsky, K. E., Halpern, J. & Johnson, J. K. 2019. The precarity of older adults living alone with cognitive impairment. *The Gerontologist,* 59**,** 271-280.

Postavaru, G.-I. 2018. A meta-ethnography of parents’ experiences of their children’s life-limiting conditions. *Qualitative Research in Psychology,* 16**,** 253-275.

Postavaru, G. I., Swaby, H. & Swaby, R. 2021. A meta-ethnographic study of fathers' experiences of caring for a child with a life-limiting illness. *Palliat Med,* 35**,** 261-279.

Price, J., Hurley, F. & Kiernan, G. 2022. ‘Managing an unexpected life - a caregiver’s career’: Parents’ experience of caring for their child with a non-malignant life-limiting condition. *Journal of Child Health Care***,** 13674935221132920.

Priddis, L., Dunwoodie, J., Balding, E., Barrett, A. & Douglas, T. 2010. Paternal experiences of their children’s diagnosis following newborn screening diagnosis. *Journal of Neonatal and Paediatric Child Health Nursing,* 13**,** 3-9.

Raina, P., O'donnell, M., Schwellnus, H., Rosenbaum, P., King, G., Brehaut, J., Russell, D., Swinton, M., King, S., Wong, M., Walter, S. D. & Wood, E. 2004. Caregiving process and caregiver burden: conceptual models to guide research and practice. *BMC pediatrics,* 4**,** 1-1.

Regmi, K., Naidoo, J. & Pilkington, P. 2010. Understanding the Processes of Translation and Transliteration in Qualitative Research. *International Journal of Qualitative Methods,* 9**,** 16-26.

Reid, A. M., Brown, J. M., Smith, J. M., Cope, A. C. & Jamieson, S. 2018. Ethical dilemmas and reflexivity in qualitative research. *Perspect Med Educ,* 7**,** 69-75.

Rigby, N. J. 2012. *Bereaved fathers' experiences of grief and mourning following the death of a child to terminal illness.*

Rivard, M. T. & Mastel-Smith, B. 2014. The lived experience of fathers whose children are diagnosed with a genetic disorder. *J Obstet Gynecol Neonatal Nurs,* 43**,** 38-49.

Robinson, J., Huskey, D., Schalley, S., Wratchford, D., Hammel, J. & Weaver, M. S. 2019a. Discovering dad: paternal roles, responsibilities, and support needs as defined by fathers of children with complex cardiac conditions perioperatively. *Cardiol Young,* 29**,** 1143-1148.

Robinson, J. E., Huskey, D., Schwartz, J. & Weaver, M. S. 2019b. The Many Roles of the Rock: A Qualitative Inquiry into the Roles and Responsibilities of Fathers of Children with Brain Tumors. *Children (Basel),* 6.

Rodriguez, A. & King, N. 2009. The lived experience of parenting a child with a life-limiting condition: a focus on the mental health realm. *Palliat Support Care,* 7**,** 7-12.

Rud, S. A., Skagestad, E. & Aasebø Hauken, M. 2023. Parents' experiences of paediatric palliative care in the community healthcare system: a qualitative study. *Palliat Care Soc Pract,* 17**,** 26323524231193036.

Schneider, M., Steele, R., Cadell, S. & Hemsworth, D. 2011. Differences on psychosocial outcomes between male and female caregivers of children with life-limiting illnesses. *J Pediatr Nurs,* 26**,** 186-99.

Schoonenboom, J. & Johnson, R. B. 2017. How to Construct a Mixed Methods Research Design. *KZfSS Kölner Zeitschrift für Soziologie und Sozialpsychologie,* 69**,** 107-131.

Schwandt, T. A. 1994. Constructivist, interpretivist approaches to human inquiry. *Handbook of qualitative research,* 1**,** 118-137.

Scott, H. M., Coombes, L., Braybrook, D., Roach, A., Harðardóttir, D., Bristowe, K., Ellis- Smith, C., Higginson, I., Gao, W., Bluebond-Langner, M., Farsides, B., Murtagh, F. E., Fraser, L. K. & Harding, R. 2022. COVID-19: Impact on Pediatric Palliative Care. *J Pain Symptom Manage,* 64**,** e1-e5.

Shardonofsky, J., Cesario, S., Fredland, N., Landrum, P., Hiatt, P. & Shardonofsky, F. 2019. The Lived Experience of Fathers Caring For a Child with Cystic Fibrosis. *Pediatric Nursing,* 45**,** 87-92.

Shaw, R. L., Booth, A., Sutton, A. J., Miller, T., Smith, J. A., Young, B., Jones, D. R. & Dixon- Woods, M. 2004. Finding qualitative research: an evaluation of search strategies. *BMC Medical Research Methodology,* 4**,** 5.

Singer, J., Roberts, K. E., Mclean, E., Fadalla, C., Coats, T., Rogers, M., Wilson, M. K., Godwin, K. & Lichtenthal, W. G. 2022. An examination and proposed definitions of family members' grief prior to the death of individuals with a life-limiting illness: A systematic review. *Palliative Medicine,* 36**,** 581-608.

Sjuls, M., Ludvigsen, M. S., Robstad, N. & Fegran, L. 2023. Fathers' experiences of living with a child with a progressive life-limiting condition without curative treatment options: A qualitative systematic review. *Journal of Advanced Nursing,* n/a.

Skamagki, G., King, A., Carpenter, C. & Wåhlin, C. 2024. The concept of integration in mixed methods research: a step-by-step guide using an example study in physiotherapy. *Physiother Theory Pract,* 40**,** 197-204.

Snaman, J. M., Feraco, A. M., Wolfe, J. & Baker, J. N. 2019. "What if?": Addressing uncertainty with families. *Pediatric Blood & Cancer,* 66**,** e27699.

Soilemezi, D. & Linceviciute, S. 2018. Synthesizing Qualitative Research. *International Journal of Qualitative Methods,* 17.

Sonnenburg, C. & Miller, Y. D. 2021. Postnatal Depression: The Role of "Good Mother" Ideals and Maternal Shame in a Community Sample of Mothers in Australia. *Sex Roles,* 85**,** 661-676.

Sörensen, S., Duberstein, P., Gill, D. & Pinquart, M. 2006. Dementia care: mental health effects, intervention strategies, and clinical implications. *The Lancet Neurology,* 5**,** 961-973.

Statistics, O. F. N. 2021. Census 2021.

Sterken, D. J. 1996. Uncertainty and coping in fathers of children with cancer. *Journal of Pediatric Oncology Nursing,* 13**,** 81-88.

Stewart, J. L. & Mishel, M. H. 2000. Uncertainty in childhood illness: a synthesis of the parent and child literature. *Sch Inq Nurs Pract,* 14**,** 299-319; discussion 321-6.

Stiawa, M., Müller-Stierlin, A., Staiger, T., Kilian, R., Becker, T., Gündel, H., Beschoner, P., Grinschgl, A., Frasch, K., Schmauß, M., Panzirsch, M., Mayer, L., Sittenberger, E. & Krumm, S. 2020. Mental health professionals view about the impact of male gender for the treatment of men with depression - a qualitative study. *BMC Psychiatry,* 20**,** 276.

Sun, D., Mao, Z., Zhang, X., Li, J. & Zhang, L. 2022. Relationship Between Post-traumatic Stress Symptoms and Anticipatory Grief in Family Caregivers of Patients With Advanced Lung Cancer: The Mediation Role of Illness Uncertainty. *Frontiers in Psychiatry,* 13.

Tan, R., Koh, S., Wong, M. E., Rui, M. & Shorey, S. 2019. Caregiver Stress, Coping Strategies, and Support Needs of Mothers Caring for their Children Who Are Undergoing Active Cancer Treatments. *Clinical Nursing Research,* 29**,** 460-468.

Taquette, S. R. & Borges Da Matta Souza, L. M. 2022. Ethical Dilemmas in Qualitative Research: A Critical Literature Review. *International Journal of Qualitative Methods,* 21**,** 16094069221078731.

Taylor, J., Booth, A., Beresford, B., Phillips, B., Wright, K. & Fraser, L. 2020. Specialist paediatric palliative care for children and young people with cancer: A mixed- methods systematic review. *Palliat Med,* 34**,** 731-775.

Teddlie, C. & Tashakkori, A. 2009. *Foundations of Mixed Methods Research: Integrating Quantitative and Qualitative Approaches in the Social and Behavioral Sciences*, SAGE Publications.

Tedeschi, R. G., Park, C. L. & Calhoun, L. G. 1998. *Posttraumatic growth: Positive changes in the aftermath of crisis*, Routledge.

Tfsl 2018. A Guide to Children's Palliative Care. Supporting babies, children and young people with life-limiting and life-threatening conditions and their families.

Tfsl 2023. Children's hospice funding in 2022/23. Together for Short Lives.

Thin, N. 2018. Qualitative approaches to culture and well-being. *In:* DIENER, E. O., S. (ed.)
Handbook of Wellbeing. NobaScholar.

Thomas, J. & Harden, A. 2008. Methods for the thematic synthesis of qualitative research in systematic reviews. *BMC Medical Research Methodology,* 8**,** 45.

Thomas, T. H., Jackson, V. A., Carlson, H., Rinaldi, S., Sousa, A., Hansen, A., Kamdar, M., Jacobsen, J., Park, E. R., Pirl, W. F., Temel, J. S. & Greer, J.

A. 2018. Communication Differences between Oncologists and Palliative Care Clinicians: A Qualitative Analysis of Early, Integrated Palliative Care in Patients with Advanced Cancer. *Journal of Palliative Medicine,* 22**,** 41-49.

Tong, A., Flemming, K., Mcinnes, E., Oliver, S. & Craig, J. 2012. Enhancing transparency in reporting the synthesis of qualitative research: ENTREQ. *BMC Medical Research Methodology,* 12**,** 181.

Tough, H., Brinkhof, M. W. G. & Fekete, C. 2020. Is informal caregiving at odds with optimal health behaviour? A cross-sectional analysis in the caregiving partners of persons with spinal cord injury. *Health Psychology and Behavioral Medicine,* 8**,** 526-542.

Toye, F., Seers, K., Allcock, N., Briggs, M., Carr, E., Andrews, J. & Barker, K. 2013. 'Trying to pin down jelly' - exploring intuitive processes in quality assessment for meta- ethnography. *BMC Medical Research Methodology,* 13**,** 46.

Tracy, S. J. 2010. Qualitative Quality: Eight "Big-Tent" Criteria for Excellent Qualitative Research. *Qualitative Inquiry,* 16**,** 837-851.

Van Grootel, L., Balachandran Nair, L., Klugkist, I. & Van Wesel, F. 2020. Quantitizing findings from qualitative studies for integration in mixed methods reviewing. *Research Synthesis Methods,* 11**,** 413-425.

Van Nes, F., Abma, T., Jonsson, H. & Deeg, D. 2010. Language differences in qualitative research: is meaning lost in translation? *European journal of ageing,* 7**,** 313-316.

Van Warmerdam, J., Zabih, V., Kurdyak, P., Sutradhar, R., Nathan, P. C. & Gupta, S. 2019. Prevalence of anxiety, depression, and posttraumatic stress disorder in parents of children with cancer: A meta-analysis. *Pediatric Blood & Cancer,* 66**,** e27677.

Vasileiou, K., Barnett, J., Barreto, M., Vines, J., Atkinson, M., Lawson, S. & Wilson, M. 2017. Experiences of Loneliness Associated with Being an Informal Caregiver: A Qualitative Investigation. *Frontiers in Psychology,* 8.

Verberne, L. M., Kars, M. C., Schouten-Van Meeteren, A. Y., Bosman, D. K., Colenbrander, D. A., Grootenhuis, M. A. & Van Delden, J. J. 2017. Aims and tasks in parental caregiving for children receiving palliative care at home: a qualitative study. *Eur J Pediatr,* 176**,** 343-354.

Verberne, L. M., Kars, M. C., Schouten-Van Meeteren, A. Y. N., Van Den Bergh, E. M. M., Bosman, D. K., Colenbrander, D. A., Grootenhuis, M. A. & Van Delden, J. J. M. 2019. Parental experiences and coping strategies when caring for a child receiving paediatric palliative care: a qualitative study. *Eur J Pediatr,* 178**,** 1075-1085.

Vernon, L., Eyles, D., Hulbert, C., Bretherton, L. & Mccarthy, M. C. 2017. Infancy and pediatric cancer: an exploratory study of parent psychological distress. *Psycho- Oncology,* 26**,** 361-368.

Vitale, D. C., Armenakis, A. A. & Feild, H. S. 2008. Integrating Qualitative and Quantitative Methods for Organizational Diagnosis: Possible Priming Effects? *Journal of Mixed Methods Research,* 2**,** 87-105.

Wallander, J. L., Varni, J. W., Babani, L., Banis, H. T., Dehaan, C. B. & Wilcox, K. T. 1989. Disability Parameters, Chronic Strain, and Adaptation of Physically Handicapped Children and Their Mothers1. *Journal of*

Pediatric Psychology, 14**,** 23-42.

Walpole, S. C. 2019. Including papers in languages other than English in systematic reviews: important, feasible, yet often omitted. *J Clin Epidemiol,* 111**,** 127-134.

Ware, J. & Raval, H. 2007. A qualitative investigation of fathers' experiences of looking after a child with a life-limiting illness, in process and in retrospect. *Clin Child Psychol Psychiatry,* 12**,** 549-65.

Whitney, C. & Evered, J. A. 2022. The Qualitative Research Distress Protocol: A Participant- Centered Tool for Navigating Distress During Data Collection. *International Journal of Qualitative Methods,* 21**,** 16094069221110317.

Who. 2011. *World Health Organisation Definition of palliative care* [Online]. http://who.int/cancer/palliative/definition/en/. [Accessed 1st December 2021].

Who. 2023. *Palliative care* [Online]. World Health Organization. Available: https://www.who.int/europe/news-room/fact-sheets/item/palliative-care [Accessed 24th October 2023].

Wills, B. S. 2009. Coping with a child with acute lymphocytic leukemia: the experiences of Chinese fathers in Hong Kong. *Cancer Nurs,* 32**,** E8-e14.

Winefield, H. R., Gill, T. K., Taylor, A. W. & Pilkington, R. M. 2012. Psychological well-being and psychological distress: is it necessary to measure both? *Psychology of Well- Being: Theory, Research and Practice,* 2**,** 3.

Wolff, J., Pak, J., Meeske, K., Worden, J. & Katz, E. 2011. Understanding Why Fathers Assume Primary Medical Caretaker Responsibilities of Children With Life-Threatening Illnesses. *Psychology of Men & Masculinity,* 12**,** 144-157.

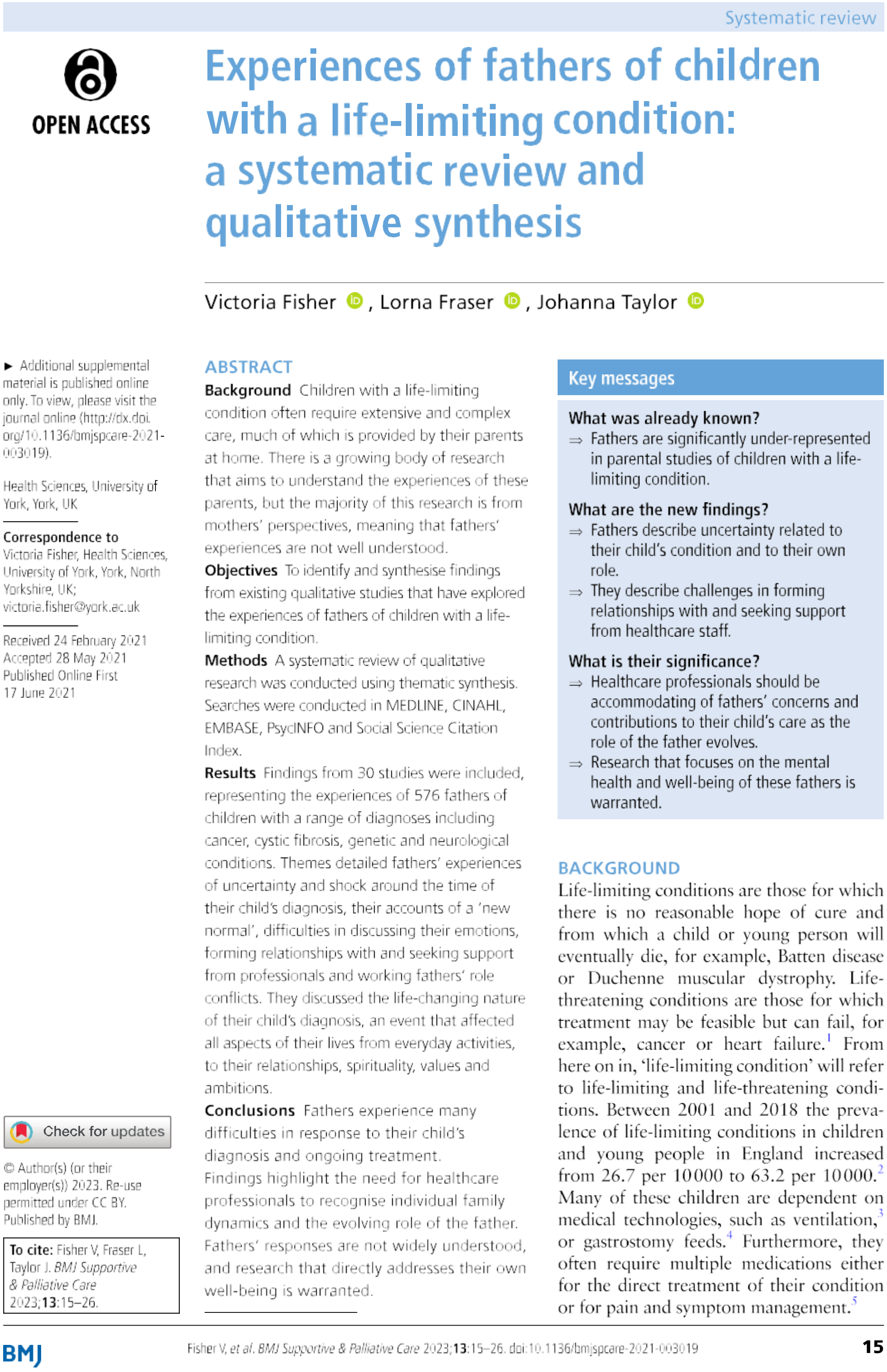
Wolff, J., Pak, J., Meeske, K., Worden, J. W. & Katz, E. 2010. Challenges and coping styles of fathers as primary medical caretakers: a multicultural qualitative study. *J Psychosoc Oncol,* 28**,** 202-17.

Woodman, C., Baillie, J. & Sivell, S. 2016. The preferences and perspectives of family caregivers towards place of care for their relatives at the end-of-life. A systematic review and thematic synthesis of the qualitative evidence. *BMJ Support Palliat Care,* 6**,** 418-429.

Yaremych, H. E. & Persky, S. 2023. Recruiting Fathers for Parenting Research: An Evaluation of Eight Recruitment Methods and an Exploration of Fathers' Motivations for Participation. *Parenting,* 23**,** 1-32.

Yu, L., Buysse, D. J., Germain, A., Moul, D. E., Stover, A., Dodds, N. E., Johnston, K. L. & Pilkonis, P. A. 2012. Development of Short Forms From the PROMIS™ Sleep Disturbance and Sleep-Related Impairment Item Banks. *Behavioral Sleep Medicine,* 10**,** 6-24.

Appendix 1: 'Experiences of fathers of children with a life-limiting condition: a systematic review and qualitative synthesis' (BMJ Supportive & Palliative Care Publication)



OPEN ACCESS

Experiences of fathers of children with a life-limiting condition: a systematic review and qualitative synthesis

Victoria Fisher, Lorna Fraser, Johanna Taylor

► Additional supplemental material is published online only. To view, please visit the journal online (http://dx.doi.org/10.1136/bmjspcare-2021-003019).

Health Sciences, University of York, York, UK

Correspondence to
Victoria Fisher, Health Sciences, University of York, York, North Yorkshire, UK; victoria.fisher@york.ac.uk

Received 24 February 2021
Accepted 28 May 2021
Published Online First 17 June 2021

To cite: Fisher V, Fraser L, Taylor J. *BMJ Supportive & Palliative Care* 2023;**13**:15–26.

ABSTRACT

Background Children with a life-limiting condition often require extensive and complex care, much of which is provided by their parents at home. There is a growing body of research that aims to understand the experiences of these parents, but the majority of this research is from mothers' perspectives, meaning that fathers' experiences are not well understood.

Objectives To identify and synthesise findings from existing qualitative studies that have explored the experiences of fathers of children with a life-limiting condition.

Methods A systematic review of qualitative research was conducted using thematic synthesis. Searches were conducted in MEDLINE, CINAHL, EMBASE, PsycINFO and Social Science Citation Index.

Results Findings from 30 studies were included, representing the experiences of 576 fathers of children with a range of diagnoses including cancer, cystic fibrosis, genetic and neurological conditions. Themes detailed fathers' experiences of uncertainty and shock around the time of their child's diagnosis, their accounts of a 'new normal', difficulties in discussing their emotions, forming relationships with and seeking support from professionals and working fathers' role conflicts. They discussed the life-changing nature of their child's diagnosis, an event that affected all aspects of their lives from everyday activities, to their relationships, spirituality, values and ambitions.

Conclusions Fathers experience many difficulties in response to their child's diagnosis and ongoing treatment. Findings highlight the need for healthcare professionals to recognise individual family dynamics and the evolving role of the father. Fathers' responses are not widely understood, and research that directly addresses their own well-being is warranted.

Key messages

What was already known?

- ⇒ Fathers are significantly under-represented in parental studies of children with a life-limiting condition.

What are the new findings?

- ⇒ Fathers describe uncertainty related to their child's condition and to their own role.
- ⇒ They describe challenges in forming relationships with and seeking support from healthcare staff.

What is their significance?

- ⇒ Healthcare professionals should be accommodating of fathers' concerns and contributions to their child's care as the role of the father evolves.
- ⇒ Research that focuses on the mental health and well-being of these fathers is warranted.

BACKGROUND

Life-limiting conditions are those for which there is no reasonable hope of cure and from which a child or young person will eventually die, for example, Batten disease or Duchenne muscular dystrophy. Life-threatening conditions are those for which treatment may be feasible but can fail, for example, cancer or heart failure.[1] From here on in, 'life-limiting condition' will refer to life-limiting and life-threatening conditions. Between 2001 and 2018 the prevalence of life-limiting conditions in children and young people in England increased from 26.7 per 10000 to 63.2 per 10000.[2] Many of these children are dependent on medical technologies, such as ventilation,[3] or gastrostomy feeds.[4] Furthermore, they often require multiple medications either for the direct treatment of their condition or for pain and symptom management.[5]

Fisher V, et al. BMJ Supportive & Palliative Care 2023;13:15–26. doi:10.1136/bmjspcare-2021-003019

Appendix 2: Medline Search Strategy

1. Fathers/
2. father*.tw.
3. dad*.tw.
4. stepfather*.tw.
5. step-father*.tw.
6. (foster* adj3 father*).tw.
7. (adopt* adj3 father*).tw.
8. 1 or 2 or 3 or 4 or 5 or 6 or 7
9. child/ or child, preschool/ or infant/
10. Adolescent/
11. (child$ or children$).tw.
12. (infant$ or infancy$).tw.
13. (baby or baby$ or babies).tw.
14. (toddler$ or kid or kids).tw.
15. (boy or boys or boyhood or girl or girls or girlhood).tw.
16. (minor or minor$ or schoolchild$).tw.
17. (adolescen$ or juvenil$ or youth$ or teen$ or "under age$" or underage$ or pubescen$).tw.
18. (pediatric$ or paediatric$ or peadiatric$).tw.
19. (young people$ or young person$).tw.
20. young adult$.tw.
21. 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19
22. life-limiting illness*.tw.
23. life-limiting condition*.tw.
24. life-limiting disease*.tw.
25. life-threatening illness*.tw.
26. life-threatening condition*.tw.
27. life-threatening disease*.tw.
28. Creutzfeldt-Jakob Syndrome/
29. (creutzfeldt-jakob$ or jakob-creutzfeldt$ or cjd or spongiform encephalopath$).ti,ab,kf.
30. Subacute Sclerosing Panencephalitis/
31. (subacute sclerosing panencephalit$ or sub-acute sclerosing panencephalit$ or sspe or subacute sclerosing leukoencephalit$ or sub-acute sclerosing leukoencephalit$ or van bogaert$ leukoencephalit$ or measles inclusion body encephalit$ or mibe).ti,ab,kf.
32. beta-Thalassemia/
33. (beta adj (thalass?emi$ or thalas?emi$)).ti,ab,kf.
34. ((thalass?emi$ or thalas?emi$) adj major).ti,ab,kf.
35. exp Anemia, Aplastic/
36. ((hypoplastic or aplastic) adj an?emi$).ti,ab,kf.
37. (medullary adj3 hypoplas$).ti,ab,kf.
38. exp Neutropenia/

39. ((severe or chronic$) adj3 neutropeni$).ti,ab,kf.
40. immunologic deficiency syndromes/ or acquired immunodeficiency syndrome/
41. (immun$ deficiency adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
42. (immunodeficiency adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
43. DiGeorge Syndrome/
44. (digeorge$ or di george$ or sedlackova$ or opitz g-bbb or velocardiofacial or velo-cardiofacial or velo-cardio-facial or shprintzen$ or ctaf).ti,ab,kf.
45. ((deletion or vcf or pharyngeal pouch or thymic aplasia or anomaly face) adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
46. Common Variable Immunodeficiency/
47. ((common variable or late onset) adj3 (immunodeficienc$ or immune deficienc$ or immunoglobulin deficienc$ or hypogammaglobulin$)).ti,ab,kf.
48. acquired hypogammaglobulin$.ti,ab,kf.
49. Cryoglobulinemia/
50. cryoglobulin?em$.ti,ab,kf.
51. Polyendocrinopathies, Autoimmune/
52. ((autoimmune or failure$) adj3 (polyglandular$ or polyendocrin$)).ti,ab,kf.
53. Progeria/
54. (progeria or hutchinson-gilford$).ti,ab,kf.
55. Tyrosinemias/
56. tyrosin?em$.ti,ab,kf.
57. Maple Syrup Urine Disease/
58. (maple syrup urine or msud).ti,ab,kf.
59. branched chain.ti,ab,kf.
60. (bckd adj5 (deficienc$ or ketoacid$ or keto-acid$)).ti,ab,kf.
61. hyperleucine-isoleucin$.ti,ab,kf.
62. Methylmalonic Acid/
63. (methylmalonic acid?emi$ or methylmalonic aciduri$ or methyl malonic acid?emi$ or methyl malonic aciduri$).ti,ab,kf.
64. Propionic Acidemia/
65. (propionic acid?em$ or propionic acidur$ or propionyl-CoA carboxylase deficienc$ or ketotic glycin?em$).ti,ab,kf.
66. Adrenoleukodystrophy/
67. (adrenoleukodystroph$ or x-ald or schilder-addison$ or addison-schilder$ or adrenomyeloneuropath$).ti,ab,kf.
68. Carnitine O-Palmitoyltransferase/
69. ((carnitine palmityltransferase or carnitine palmitoyltransferase or carnitine o-palmityltransferase or carnitine o-palmitoyltransferase) adj3 deficienc$).ti,ab,kf.
70. Fanconi Syndrome/
71. (fanconi$ adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
72. Cystinosis/
73. (cystinos$ or cystine storage or cystine diathes$ or cystine disease$).ti,ab,kf.
74. Oculocerebrorenal Syndrome/

75. ((lowe or lowes or oculocerebrorenal or cerebrooculorenal or cerebro-oculo-renal) adj3 (syndrome$ or disease$ or disorder$)).ti,ab,kf.
76. Metalloproteins/df [Deficiency]
77. Molybdenum/df [Deficiency]
78. (molybdenum cofactor deficien$ or molybdenum co-factor deficien$).ti,ab,kf.
79. Oxidoreductases Acting on Sulfur Group Donors/df [Deficiency]
80. Sulfite Oxidase/df [Deficiency]
81. ((sulphite$ or sulfite$) adj3 oxidase deficien$).ti,ab,kf.
82. Argininosuccinic Acid/
83. (argininosuccinic acidur$ or argininosuccinic acid?emi$).ti,ab,kf.
84. Citrullinemia/
85. (citrullin?emi$ or citrullinuri$).ti,ab,kf.
86. Amino Acid Metabolism, Inborn Errors/
87. (glutaric acid?emi$ or glutaric aciduri$).ti,ab,kf.
88. Hyperglycinemia, Nonketotic/
89. (glycine encephalopath$ or non-ketotic hyperglycin?emi$ or nonketotic hyperglycin?emi$).ti,ab,kf.
90. Hyperargininemia/
91. (arginin?emi$ or arginase deficien$ or hyperarginin?emi$).ti,ab,kf.
92. Renal Aminoacidurias/
93. (aminoaciduri$ or aminoacid?emi$).ti,ab,kf.
94. exp glycogen storage disease/
95. (glycogen storage adj (disease$ or syndrome$ or disorder$)).ti,ab,kf.
96. (pompe$ adj (disease$ or syndrome$ or disorder$)).ti,ab,kf.
97. Galactosemias/
98. galactos?emi$.ti,ab,kf.
99. Pyruvate Dehydrogenase Complex Deficiency Disease/
100. (pyruvate dehydrogenase adj3 deficien$).ti,ab,kf.
101. (oxalosis and (renal or kidney$)).ti,ab,kf.
102. exp Gangliosidoses/
103. gangliosidos$.ti,ab,kf.
104. (sandhoff$ adj (disease$ or syndrome$ or disorder$)).ti,ab,kf.
105. tay sach$.ti,ab,kf.
106. Mucolipidoses/
107. mucolipidos$.ti,ab,kf.
108. Canavan Disease/
109. (canavan$ leucodystroph$ or aspartoacylase deficien$ or aminoacylase 2 deficien$).ti,ab,kf.
110. ((canavan$ or canavan-van bogaert-bertrand$) adj (disease$ or syndrome$ or disorder$)).ti,ab,kf.
111. Gaucher Disease/
112. (gaucher$ adj (disease$ or syndrome$ or disorder$)).ti,ab,kf.
113. (glucocerebrosidase deficien$ or glucosylceramidase deficien$).ti,ab,kf.
114. Leukodystrophy, Metachromatic/

115. (metachromatic leukodystroph$ or arylsulfatase A deficien$ or metachromic leukodystroph$).ti,ab,kf.
116. exp Niemann-Pick Diseases/
117. (niemann-pick$ or sphingomyelinase deficien$).ti,ab,kf.
118. Sphingolipidoses/
119. sphingolipidos$.ti,ab,kf.
120. Fabry Disease/
121. (fabry$ adj (disease$ or syndrome$ or disorder$)).ti,ab,kf.
122. (angiokeratoma corporis diffusum or alpha-galactosidase A deficien$).ti,ab,kf.
123. Leukodystrophy, Globoid Cell/
124. (krabbe$ adj (disease$ or syndrome$ or disorder$)).ti,ab,kf.
125. (globoid cell leukodystroph$ or galactosylceramide lipidos$ or galactosylcerebrosidase deficien$ or galactosylceramidase deficien$).ti,ab,kf.
126. Farber Lipogranulomatosis/
127. (farber$ adj (disease$ or syndrome$ or disorder$)).ti,ab,kf.
128. (farber$ lipogranulomatos$ or ceramidase deficien$ or fibrocytic dysmucopolysaccharidos$).ti,ab,kf.
129. Pelizaeus-Merzbacher Disease/
130. pelizaeus-merzbacher$.ti,ab,kf.
131. Sulfatases/df [Deficiency]
132. Multiple Sulfatase Deficiency Disease/
133. (sulfatase deficien$ or sulphatase deficien$ or mucosulfatidos$).ti,ab,kf.
134. (austin$ adj (disease$ or syndrome$ or disorder$)).ti,ab,kf.
135. sulfatidos$.ti,ab,kf.
136. Sea-Blue Histiocyte Syndrome/
137. sea-blue histiocyt$.ti,ab,kf.
138. Neuronal Ceroid-Lipofuscinoses/
139. (batten$ adj (disease$ or syndrome$ or disorder$)).ti,ab,kf.
140. (neuronal ceroid lipofuscinos$ or santavuori-haltia$ or jansky-bielschowsky$ or bielschowsky-jansky$).ti,ab,kf.
141. (kuf$ adj (disease$ or syndrome$ or disorder$)).ti,ab,kf.
142. spielmeyer vogt$.ti,ab,kf.
143. Xanthomatosis, Cerebrotendinous/
144. ((cerebrotendineous or cerebrotendinous or cerebrotendious or cerebral) adj3 (xanthomatos$ or cholesteros$)).ti,ab,kf.
145. bogaert-scherer-epstein$.ti,ab,kf.
146. Wolman Disease/
147. (wolman$ adj (disease$ or syndrome$ or disorder$)).ti,ab,kf.
148. lysosomal acid lipase deficien$.ti,ab,kf.
149. exp Mucopolysaccharidoses/
150. mucopolysaccharidos$.ti,ab,kf.
151. (hurler$ adj2 (syndrome$ or disease$ or disorder$)).ti,ab,kf.
152. (hunter$ adj2 (syndrome$ or disease$ or disorder$)).ti,ab,kf.

153. (MPS1 or MPS2 or MPS3 or MPS4 or MPS5 or MPS6 or MPS7 or MPS-1 or MPS-2 or MPS-3 or MPS-4 or MPS-5 or MPS-6 or MPS-7 or MPSI or MPSII or MPSIII or MPSIV or MPSV or MPSVI or MPSVII or MPS-I or MPS-II or MPS-III or MPS-IV or MPS-V or MPS-VI or MPS-VII).ti,ab,kf.
154. (beta glucuronidase deficien$ or sly syndrome$ or sly disorder$ or sly disease$).ti,ab,kf.
155. (maroteaux-lamy$ or marotaeux-lamy$ or polydystrophic dwarfism).ti,ab,kf.
156. (morquio$ or moriquio$ or beta galactosidase deficien$).ti,ab,kf.
157. (sanfilippo$ or sanfillipo$).ti,ab,kf.
158. Mucolipidoses/
159. (mucolipidos$ or pseudo-hurler$ or pseudohurler$).ti,ab,kf.
160. ((inclusion-cell or i-cell) adj (disease$ or syndrome$ or disorder$)).ti,ab,kf.
161. Fucosidosis/
162. (fucosidos$ or fucidos$).ti,ab,kf.
163. "Congenital Disorders of Glycosylation"/
164. ((cdg or ctg) adj (disease$ or disorder$ or syndrome$)).ti,ab,kf.
165. (carbohydrate-deficient glycoprotein adj (disease$ or disorder$ or syndrome$)).ti,ab,kf.
166. (congenital disorder$ adj3 glycosylation).ti,ab,kf.
167. Lesch-Nyhan Syndrome/
168. ((nyhan$ or kelley-seegmiller$) adj (syndrome$ or disorder$ or disease$)).ti,ab,kf.
169. juvenile gout.ti,ab,kf.
170. Menkes Kinky Hair Syndrome/
171. menkes$.ti,ab,kf.
172. ((copper transport or steely hair or kinky hair) adj (disease$ or syndrome$ or disorder$)).ti,ab,kf.
173. alpha 1-Antitrypsin Deficiency/
174. (antitrypsin deficien$ or A1AD).ti,ab,kf.
175. (AAT deficien$ or alpha-1 protease deficien$).ti,ab,kf.
176. bisalbumin?emi$.ti,ab,kf.
177. Lipodystrophy, Congenital Generalized/
178. (congenital generali?ed lipodystroph$ or berardinelli$ or bernardnelli$).ti,ab,kf.
179. Landau-Kleffner Syndrome/
180. (landau-kleffner$ or infantile acquired aphasia$ or acquired epileptic aphasia$).ti,ab,kf.
181. (aphasia$ adj5 convulsive).ti,ab,kf.
182. Rett Syndrome/
183. (rett$ adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
184. cerebroatrophic hyperammon?emi$.ti,ab,kf.
185. Huntington Disease/
186. huntington$.ti,ab,kf.
187. exp Spinocerebellar Ataxias/

188. (spinocerebellar ataxia$ or ataxia$ telangiectasia$ or louis-bar$ syndrome$ or louis-bar$ disease$ or louis-bar$ disorder$ or machado-joseph$ or joseph$ disease$ or joseph$ disorder$ or joseph$ syndrome$).ti,ab,kf.
189. Friedreich Ataxia/
190. ((friedreich$ or friedrich$) adj3 ataxia$).ti,ab,kf.
191. spinocerebellar degenerat$.ti,ab,kf.
192. "Spinal Muscular Atrophies of Childhood"/
193. (spinal muscular atroph$ or werdnig hoffman$).ti,ab,kf.
194. (dubowitz$ or kugelberg-welander$).ti,ab,kf.
195. Bulbar Palsy, Progressive/
196. (fazio-londe$ or faziolonde$ or progressive bulbar pals$).ti,ab,kf.
197. Pantothenate Kinase-Associated Neurodegeneration/
198. (pantothenate kinase-associated neurodegenerat$ or PKAN or hallervorden-spatz$).ti,ab,kf.
199. ((neurodegeneration adj3 brain iron accumulation) or NBIA$1).ti,ab,kf.
200. Olivopontocerebellar Atrophies/
201. (olivopontocerebellar atroph$ or OPCA or olivopontocerebellar degenerat$).ti,ab,kf.
202. (multiple system atrophy adj5 cerebellar).ti,ab,kf.
203. "Diffuse Cerebral Sclerosis of Schilder"/
204. (alper$ adj (disease$ or syndrome$ or disorder$)).ti,ab,kf.
205. (progressive sclerosing poliodystroph$ or progressive infantile poliodystroph$).ti,ab,kf.
206. (diffuse cerebral sclerosis adj5 schilder$).ti,ab,kf.
207. Leigh Disease/
208. (leigh$ adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
209. (subacute necrotizing encephalomyelopath$ or subacute necrotising encephalomyelopath$ or sub-acute necrotizing encephalomyelopath$ or sub-acute necrotising encephalomyelopath$ or SNEM).ti,ab,kf.
210. (aicardi-gouti?res or aicardia-gouti?res).ti,ab,kf.
211. (worster-drought$ or congenital suprabulbar pares$).ti,ab,kf.
212. multiple sclerosis/ or multiple sclerosis, chronic progressive/ or multiple sclerosis, relapsing-remitting.mp.
213. (multiple sclerosis or disseminated sclerosis or encephalomyelitis disseminata$).ti,ab,kf.
214. (demyelinating adj (disease$ or syndrome$ or disorder$)).ti,ab,kf.
215. exp Epilepsies, Myoclonic/
216. myoclonic epileps$.ti,ab,kf.
217. ((lafora$ or merrf$ or unverricht-lundborg$ or janz$) adj (disease$ or syndrome$ or disorder$)).ti,ab,kf.
218. lennox-gastaut$.ti,ab,kf.
219. (lennox$ adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
220. Spasms, Infantile/
221. (west$ adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
222. Epilepsia Partialis Continua/

223. (epilepsia partialis continua or kojevnikov$ or epilepsia partialis continuoa or kozhevnikof$).ti,ab,kf.
224. Charcot-Marie-Tooth Disease/
225. (charcot-marie-tooth$ or peroneal muscular atroph$).ti,ab,kf.
226. (progressive neuropathic muscular atroph$ or hereditary peroneal nerve dysfunction$ or peroneal neuropath$).ti,ab,kf.
227. "Hereditary Sensory and Motor Neuropathy"/
228. (hereditary sensory adj3 motor neuropath$).ti,ab,kf.
229. (hereditary motor adj3 sensory neuropath$).ti,ab,kf.
230. Refsum Disease, Infantile/
231. Peroxisomal Disorders/
232. (infantile refsum or infantile phytanic acid storage).ti,ab,kf.
233. Myasthenic Syndromes, Congenital/
234. congenital myasth?eni$.ti,ab,kf.
235. Muscular Dystrophy, Duchenne/
236. (duchenne muscular dystroph$ or dmd).ti,ab,kf.
237. exp Muscular Dystrophies, Limb-Girdle/
238. (limb-girdle or erb$ muscular dystroph$).ti,ab,kf.
239. (sarcoglycanopath$ or sarcoglycaopath$).ti,ab,kf.
240. Osteochondrodysplasias/
241. (osteochondrodysplas$ or schwartz-jampel or chondrodystrophi$ myotoni$ or myotoni$ chondrodystrophi$).ti,ab,kf.
242. Myotonia Congenita/
243. (congenita$ myotoni$ or myotoni$ congenita$).ti,ab,kf.
244. (thomsen$ adj (disease$ or disorder$ or syndrome$)).ti,ab,kf.
245. ((recessive adj3 myotoni$) or becker$ myotoni$).ti,ab,kf.
246. Isaacs Syndrome/
247. (isaac$ adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
248. neuromyotoni$.ti,ab,kf.
249. Myotonic Disorders/
250. (paramyotoni$ congenita$ or congenita$ paramyotoni$).ti,ab,kf.
251. (eulenburg$ adj (disease$ or syndrome$ or disorder$)).ti,ab,kf.
252. (myotoni$ adj (disease$ or disorder$ or syndrome$)).ti,ab,kf.
253. pseudomyotoni$.ti,ab,kf.
254. exp Myopathies, Structural, Congenital/
255. (congenital adj3 myopath$).ti,ab,kf.
256. myopathycongenital.ti,ab,kf.
257. ((nemaline or rod) adj3 myopath$).ti,ab,kf.
258. ((central core or mini-core or minicore or multicore or multi-core) adj (disease$ or disorder$ or syndrome$ or myopath$)).ti,ab,kf.
259. fiber type disproportion.ti,ab,kf.
260. fibre type disproportion.ti,ab,kf.
261. Muscular Dystrophies/cn [Congenital]
262. (congenital$ adj5 muscular dystroph$).ti,ab,kf.

263. ((centronuclear or myotubular) adj myopath$).ti,ab,kf.
264. exp Mitochondrial Myopathies/
265. (mitochondrial myopath$ or mitochondrial encephalomyopath$ or chronic progressive external ophthalmopleg$).ti,ab,kf.
266. ((melas or kearns-sayre$) adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
267. Quadriplegia/ and spastic$.ti,ab,kf.
268. (spastic quadriplegi$ or spastic tetraplegi$).ti,ab,kf.
269. Reye Syndrome/
270. (reye$ adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
271. multiple pterygium.ti,ab,kf.
272. Hypertension, Pulmonary/ and primary$.ti,ab,kf.
273. ((primary pulmonary or precapillary pulmonary or idiopathic pulmonary) adj (hypertension or ht or arterial hypertension)).ti,ab,kf.
274. ((primary bronchopulmonary or precapillary bronchopulmonary or idiopathic bronchopulmonary) adj (hypertension or ht or arterial hypertension)).ti,ab,kf.
275. ((primary lung or precapillary lung or idiopathic lung) adj (hypertension or ht or arterial hypertension)).ti,ab,kf.
276. ipah.ti,ab,kf.
277. Cardiomyopathy, Dilated/
278. ((congestive or dilated) adj cardiomyopath$).ti,ab,kf.
279. exp Cardiomyopathy, Hypertrophic/
280. (hypertrophic adj cardiomyopath$).ti,ab,kf.
281. Cardiomyopathies/cn [Congenital]
282. (congenital adj3 cardiomyopath$).ti,ab,kf.
283. Cardiomyopathy, Restrictive/
284. (restrictive cardiomyopath$ or obliterative cardiomyopath$ or constrictive cardiomyopath$).ti,ab,kf.
285. exp Pulmonary Fibrosis/
286. (pulmonary fibros$ or lung fibros$ or bronchopulmonary fibros$ or fibrosing alveolit$ or interstitial pneumonit$).ti,ab,kf.
287. Respiratory Insufficiency/
288. (respiratory adj (failure$ or insufficienc$)).ti,ab,kf.
289. "Cystic Adenomatoid Malformation of Lung, Congenital"/
290. ((cystic lung or cystic pulmonary or cystic bronchopulmonary) adj (disease$ or disorder or syndrome$)).ti,ab,kf.
291. (bronchogenic cyst$ or bronchopulmonary foregut malformation$).ti,ab,kf.
292. cystic adenomatoid malformation$.ti,ab,kf.
293. lobar emphysem$.ti,ab,kf.
294. (pulmonary sequestration$ or bronchopulmonary sequestration$ or lung sequestration$ or extralobar sequestration$ or extra-lobar sequestration$ or intralobar sequestration$ or intra-lobar sequestration$).ti,ab,kf.
295. pulmolithias$.ti,ab,kf.
296. exp Liver Failure/
297. ((liver$1 or hepatic) adj3 fail$).ti,ab,kf.

298. exp Liver Cirrhosis/
299. (cirrhosis adj3 liver$1).ti,ab,kf.
300. Hepatic Veno-Occlusive Disease/
301. ((veno-occlusive or venous occlusive) adj (disease$ or syndrome$ or disorder$)).ti,ab,kf.
302. Exocrine Pancreatic Insufficiency/
303. (swachman-diamond or shwachman-bodian or schwachmann-diamond or shwachmann-bodian).ti,ab,kf.
304. Granulomatosis with Polyangiitis/
305. wegener$ granulomatos$.ti,ab,kf.
306. (granulomatos$ adj3 polyangiit$).ti,ab,kf.
307. Osteolysis, Essential/
308. essential osteolys$.ti,ab,kf.
309. ((gorham$ or gorham-stout$ or vanishing bone or phantom bone) adj (disease$ or syndrome$ or disorder)).ti,ab,kf.
310. ((arc or arthrogryposis renal dysfunction cholestasis) adj (disease$ or syndrome$ or disorder)).ti,ab,kf.
311. Cerebral Hemorrhage/cn [Congenital]
312. Cerebral Hemorrhage, Traumatic/
313. Cerebral Hemorrhage/ and Birth Injuries.mp.
314. (cerebral h?emorrhage$ and (birth$ adj3 injur$)).ti,ab,kf.
315. Asphyxia Neonatorum/
316. asphyxia neonatorum.ti,ab,kf.
317. ((perinatal$ or neonatal$ or birth$) adj3 asphyxia$).ti,ab,kf.
318. Rubella Syndrome, Congenital/
319. congenital rubella.ti,ab,kf.
320. exp Cytomegalovirus Infections/cn [Congenital]
321. (congenital adj (cytomegalovirus$ or cmv)).ti,ab,kf.
322. Chickenpox/cn [Congenital]
323. exp Herpes Zoster/cn [Congenital]
324. Herpesvirus 3, Human/ and congenital$.ti,ab,kf.
325. ((congenital or fetal or foetal) adj3 (varicella$ or chicken pox$ or VZV)).ti,ab,kf.
326. Toxoplasmosis, Congenital/
327. congenital toxoplasmos$.ti,ab,kf.
328. exp Hypoxia, Brain/
329. ((brain$ or cerebral) adj3 hypoxi$).ti,ab,kf.
330. Renal Insufficiency/cn [Congenital]
331. Acute Kidney Injury/cn [Congenital]
332. Renal Insufficiency, Chronic/cn [Congenital]
333. Kidney Failure, Chronic/cn [Congenital]
334. (congenital$ adj3 (kidney failure$ or renal failure$ or kidney insufficienc$ or renal insufficienc$)).ti,ab,kf.
335. (congenital$ adj3 (kidney disease$ or renal disease$)).ti,ab,kf.
336. Anencephaly/

337. (anencephal$ or meroanencephal$ or craniorachischis$).ti,ab,kf.
338. (aprosencephal$ adj3 open cranium).ti,ab,kf.
339. Encephalocele/
340. (encephalocele$ or cranium bifidum).ti,ab,kf.
341. Dandy-Walker Syndrome/
342. dandy-walker$.ti,ab,kf.
343. Acrocallosal Syndrome/
344. (acrocallosal or acro-callosal or acrocolossal or acro colossal).ti,ab,kf.
345. Aicardi Syndrome/
346. (aicardi$ adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
347. Holoprosencephaly/
348. (holoprosencephal$ or arhinencephal$ or holosprosencephal$).ti,ab,kf.
349. Hydranencephaly/
350. (hydranencephal$ or hydrancephal$ or hydroanencephal$).ti,ab,kf.
351. exp Lissencephaly/
352. Microcephaly/
353. (lissencephal$ or walker-warburg$ or miller-dieker$ or norman-robert$ or microlissencephal$).ti,ab,kf.
354. ((fukuyama$ or muscle-eye-brain) adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
355. "Malformations of Cortical Development"/
356. (microgyria$ or microgyrus or micro-gyria$ or micro-gyrus).ti,ab,kf.
357. (pachygyria$ or pachgyria$).ti,ab,kf.
358. agyria$.ti,ab,kf.
359. Septo-Optic Dysplasia/
360. ((septo-optic or septooptic) adj dysplas$).ti,ab,kf.
361. de morsier$.ti,ab,kf.
362. (schizencephal$ or schizzencephal$).ti,ab,kf.
363. Arnold-Chiari Malformation/
364. chiari$ malformation$.ti,ab,kf.
365. Truncus Arteriosus, Persistent/
366. (truncus or common arterial trunk$).ti,ab,kf.
367. "Transposition of Great Vessels"/
368. ((transposition$ or dextrotransposition$ or dtransposition$ or levotransposition$ or ltransposition$) adj3 (great arter$ or main arter$ or aorta$ or pulmonary arter$ or great vessel$ or main vessel$)).ti,ab,kf.
369. (dextro-tga or d-tga or levo-tga or l-tga).ti,ab,kf.
370. (double inlet adj3 ventricle$).ti,ab,kf.
371. DILV.ti,ab,kf.
372. single ventricle$.ti,ab,kf.

373. Heart Defects, Congenital/ and Atrial Appendage.mp.
374. (isomerism adj3 atrial appendage$).ti,ab,kf.
375. (aspleni$ or polyspleni$ or poly-spleni$).ti,ab,kf.

376. "Tetralogy of Fallot"/
377. (tetralogy adj3 fallot$).ti,ab,kf.
378. Eisenmenger Complex/
379. (eisenmenger$ or tardive cyanos$ or eisenmeyer$).ti,ab,kf.
380. (pentalogy adj3 fallot$).ti,ab,kf.
381. Pulmonary Atresia/
382. ((pulmonary or bronchopulmonary or lung$) adj3 atresia$).ti,ab,kf.
383. Tricuspid Atresia/
384. ((tricuspid or tri) adj3 atresia$).ti,ab,kf.
385. Ebstein Anomaly/
386. (ebstein$ adj (anomal$ or malformation$)).ti,ab,kf.
387. Hypoplastic Left Heart Syndrome/
388. (hypoplastic left heart adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
389. ((aortic or aorta$) adj3 atresia$).ti,ab,kf.
390. (mitral adj3 atresia$).ti,ab,kf.
391. ((absence$ or absent$) adj3 (aorta$ or aortic)).ti,ab,kf.
392. (aplas$ adj3 (aorta$ or aortic)).ti,ab,kf.
393. exp Aortic Aneurysm/cn [Congenital]
394. (((aorta$ or aortic) adj3 aneurys$) and congenital$).ti,ab,kf.
395. (hypoplas$ adj3 (aorta$ or aortic)).ti,ab,kf.
396. (convulsion$ adj3 (aorta$ or aortic)).ti,ab,kf.
397. (persistent right adj3 (aorta$ or aortic)).ti,ab,kf.
398. ((anomalous pulmonary venous or anamolous pulmonary venous) adj (connection or drainage or return)).ti,ab,kf.
399. ((absence$ or absent$) adj3 vena$ cava$).ti,ab,kf.
400. (persistent left adj3 cardinal vein$).ti,ab,kf.
401. Scimitar Syndrome/
402. ((scimitar$ or pulmonary venolobar) adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
403. (arteriovenous malformations/ or intracranial arteriovenous malformations/) and bilateral.ti,ab,kf.
404. ((bilateral AV or bilateral arteriovenous or bilateral arterio-venous) adj3 malform$).ti,ab,kf.
405. ((trachea$ or windpipe$ or wind-pipe$) adj3 atresia$).ti,ab,kf.
406. Tracheal Stenosis/
407. ((trachea$ or laryngotrachea$ or glottic or subglottic or sub-glottic) adj3 stenosis).ti,ab,kf.
408. Bronchopulmonary Dysplasia/
409. ((lung$ or pulmonary or bronchopulmonary) adj3 (hypoplas$ or dysplas$)).ti,ab,kf.
410. ((absence$ or absent$) adj3 (esophag$ or oesophag$ or foodpipe or food-pipe$ or gullet$)).ti,ab,kf.
411. Intestinal Atresia/
412. (duoden$ adj3 atresia$).ti,ab,kf.
413. ((absence$ or absent$) adj3 (intestin$ or gastrointestin$)).ti,ab,kf.

414. ((intestin$ or gastrointestin$) adj3 atresia$).ti,ab,kf.
415. ((intestin$ or gastrointestin$) adj3 stenos$).ti,ab,kf.
416. (cloaca$ adj3 (abnor$ or malform$ or anomal$)).ti,ab,kf.
417. (cloaca$ adj3 exopthalmo$).ti,ab,kf.
418. Biliary Atresia/
419. (biliary adj3 atresia$).ti,ab,kf.
420. (extrahepatic ductopen$ or extra-hepatic ductopen$ or progressive obliterative cholangiopath$).ti,ab,kf.
421. (biliary adj3 hypoplas$).ti,ab,kf.
422. (alagille$ adj3 atresia$).ti,ab,kf.
423. ((absence$ or absent$) adj3 kidney$).ti,ab,kf.
424. (potter$ adj (sequence$ or syndrome$ or disease$ or disorder$)).ti,ab,kf.
425. Oligohydramnios/
426. oligohydramn$.ti,ab,kf.
427. Multicystic Dysplastic Kidney/
428. ((kidney$ or renal) adj3 dysplas$).ti,ab,kf.
429. ((meckel$ or meckelgruber$ or gruber$) adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
430. dysencephalia splanchnocystica$.ti,ab,kf.
431. (pena-shokeir$ or penn-shokeir$).ti,ab,kf.
432. (larsen$ adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
433. Acrocephalosyndactylia/
434. acrocephalosyndactyl$.ti,ab,kf.
435. (pfeiffer$ adj (syndrome$ or disease$ or syndrome$)).ti,ab,kf.
436. Short Rib-Polydactyly Syndrome/
437. short rib$1.ti,ab,kf.
438. (saldino-noonan$ or majewski$ or verma-naumoff$ or beemer-langer$).ti,ab,kf.
439. (jeune$ adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
440. asphyxiating thoracic dysplas$.ti,ab,kf.
441. exp Chondrodysplasia Punctata/
442. chondrodysplasia punctata$.ti,ab,kf.
443. ((conradi$ or h?nermann$ or happle$) adj3 (syndrome$ or disease$ or disorder$)).ti,ab,kf.
444. Osteogenesis Imperfecta/
445. osteogenesis imperfecta.ti,ab,kf.
446. ((brittle bone or lobstein$) adj (disease$ or disorder$ or syndrome$)).ti,ab,kf.
447. Osteochondrodysplasias/
448. (spondyloepimetaphyseal or spondyloepiphyseal or spendylo metaphyseal).ti,ab,kf.
449. Hernia, Umbilical/
450. (omphalocele$ or omphalocoele$ or exomphalos).ti,ab,kf.
451. (hernia$ adj3 umbilic$).ti,ab,kf.
452. Gastroschisis/
453. gastroschis$.ti,ab,kf.

454. Ichthyosis, Lamellar/
455. (lamellar$ adj3 ichthyos$).ti,ab,kf.
456. ((harlequin$ or harloquin$) adj3 (ichthyos$ or baby or babies or f?etus$)).ti,ab,kf.
457. (ichthyosis congenita$ or ichthyosis fetalis or keratosis diffusa fetalis).ti,ab,kf.
458. exp Epidermolysis Bullosa/
459. epidermolysis bullosa$.ti,ab,kf.
460. (johanson-blizzard$ or johanna-blizzard$).ti,ab,kf.
461. Xeroderma Pigmentosum/
462. xeroderma pigmentosum.ti,ab,kf.
463. Ectodermal Dysplasia/
464. lacrimo-auriculo-dento-digital.ti,ab,kf.
465. ectodermal dysplas$.ti,ab,kf.
466. ((ladd or eec) adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
467. Sturge-Weber Syndrome/
468. (sturge-weber or encephalotrigeminal angiomatos$).ti,ab,kf.
469. Fetal Alcohol Spectrum Disorders/
470. f?etal alcohol.ti,ab,kf.
471. Pierre Robin Syndrome/
472. pierre robin$.ti,ab,kf.
473. Acrocephalosyndactylia/
474. (acrocephalosyndact$ or acrocephalopolysyndact$).ti,ab,kf.
475. ((apert$ or crouzon$ or saethre-chotzen$ or noack$ or carpenter$ or sakati-nyhan-tisdale$ or goodman$) adj (syndrome$ or disorder$ or disease$)).ti,ab,kf.
476. Fraser Syndrome/
477. (fraser$ adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
478. cryptophthalmos.ti,ab,kf.
479. (cyclopia$1 or cyclocephal$ or synophthalmi$).ti,ab,kf.
480. Goldenhar Syndrome/
481. (goldenhar$ or oculo-auriculo-vertebral).ti,ab,kf.
482. Mobius Syndrome/
483. ((m?bius$ or moebius$) adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
484. Orofaciodigital Syndromes/
485. (orofaciodigital or oro-facial-digital or oral-facial-digital or papillon-league$ or psaume$).ti,ab,kf.
486. (robin$ adj (syndrome$ or disorder$ or disease$)).ti,ab,kf.
487. (freeman-sheldon$ or distal arthrogrypos$ or craniocarpotarsal dysplas$ or craniocarpotarsal dystroph$ or canio-carpo-tarsal or windmill-vane-hand$ or whistling-face).ti,ab,kf.
488. De Lange Syndrome/
489. ((de lange$ or bushy$) adj (syndrome$ or disorder$ or disease$)).ti,ab,kf.
490. amsterdam dwarfism.ti,ab,kf.
491. (aarskog or faciodigitogenital or facio-digito-genital or facial digital genital or shawl scrotum or faciogenital or facio-genital).ti,ab,kf.
492. Cockayne Syndrome/

493. (cockayne$ or neill-dingwall$).ti,ab,kf.
494. (cerebro-oculo-facio-skeletal or cerebro-oculo-facial-skeletal).ti,ab,kf.
495. (dubowitz$ adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
496. (robinow$ or robinhow$).ti,ab,kf.
497. (f?etal face or f?etal facies or f?etal faces or acral dysostos$ or mesomelic dwarfism or covesdem$).ti,ab,kf.
498. Silver-Russell Syndrome/
499. (silver-russell$ or russell-silver$).ti,ab,kf.
500. (silver$ adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
501. ((seckel$ or harper$) adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
502. (microcephalic primordial dwarfism or bird-headed dwarf$ or virchow-seckel dwarfism).ti,ab,kf.
503. Smith-Lemli-Opitz Syndrome/
504. (smith-lemli-opitz$ or dehydrocholesterol reductase deficien$).ti,ab,kf.
505. Prader-Willi Syndrome/
506. (prader-willi$ or pradar-willi$).ti,ab,kf.
507. Rubinstein-Taybi Syndrome/
508. (rubinstein-taybi$ or rubenstein-tabyii$ or broad thumb-hallux).ti,ab,kf.
509. ((rubinstein$ or rubenstein$) adj2 (syndrome$ or disease$ or disorder$)).ti,ab,kf.
510. Nephritis, Hereditary/
511. (alport$ adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
512. (hereditary nephritis or h?emorrhagic familial nephritis).ti,ab,kf.
513. (hereditary deafness adj3 nephropath$).ti,ab,kf.
514. (h?ematuria adj3 nephropath$ adj3 deafness).ti,ab,kf.
515. Laurence-Moon Syndrome/
516. laurence-moon$.ti,ab,kf.
517. Bardet-Biedl Syndrome/
518. (bardet-biedl$ or biedl-bardet$).ti,ab,kf.
519. Zellweger Syndrome/
520. zellweger$.ti,ab,kf.
521. ((cerebrohepatorenal or cerebro-hepato-renal) adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
522. (edward$ adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
523. "trisomy 18".ti,ab,kf.
524. (patau$ adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
525. ("trisomy 13" or "trisomy D").ti,ab,kf.
526. "trisomy 22".ti,ab,kf.
527. "trisomy 9".ti,ab,kf.
528. "trisomy 10".ti,ab,kf.
529. duplication syndrome$.ti,ab,kf.
530. (("chromosome 8" or "chr 8") adj5 duplicat$).ti,ab,kf.
531. Chromosome Duplication/
532. exp X Chromosome/ab [Abnormalities]
533. exp X Chromosome/ and duplicat$.ti,ab,kf.

534. (("chromosome x" or "chr x") and duplicat$).ti,ab,kf.
535. (chromosom$ abnormality adj5 duplicat$).ti,ab,kf.
536. "tetrasomy 5p".ti,ab,kf.
537. (tetrasomy adj3 mosaic$).ti,ab,kf.
538. Chromosomes, Human, Pair 5/ and Mosaicism.mp.
539. Tetrasomy/
540. Trisomy/ and (chromosomes, human, pair 9/ or chromosomes, human, pair 10/ or chromosomes, human, pair 13/ or Chromosomes, Human, Pair 18/ or chromosomes, human, pair 22/)
541. Chromosome Deletion/ and Chromosomes, Human, Pair 4/
542. (delet$ adj5 short arm adj5 "chrom$ 4").ti,ab,kf.
543. Wolf-Hirschhorn Syndrome/
544. ((wolf-hirschhorn$ or wolff hirschorn$ or chromosome deletion dillan$ or pitt-rogers-dank$ or pitt$) adj3 (syndrome$ or disease$ or disorder$)).ti,ab,kf.
545. Cri-du-Chat Syndrome/
546. ((cri du chat$ or crying cat$ or 5p or lejeune$) adj3 (syndrome$ or disease$ or disorder$)).ti,ab,kf.
547. Jacobsen Distal 11q Deletion Syndrome/
548. ((jacobsen$ or 11q deletion) adj5 (syndrome$ or disease$ or disorder$)).ti,ab,kf.
549. exp Monosomy/ and Chromosomes, Human, Pair 9/
550. (9p minus or 9p deletion).ti,ab,kf.
551. (alfi$ adj (syndrome$ or disease$ or disorder$)).ti,ab,kf.
552. (degouchy$ or de gouchy$ or degrouchy$ or de grouchy$).ti,ab,kf.
553. distal 18q.ti,ab,kf.
554. Hypoventilation/cn [Congenital]
555. (ondine$ curse or congenital central hypoventilation or primary alveolar hypoventilation).ti,ab,kf.
556. Graft vs Host Disease/ and (Chronic Disease/ or chronic$.ti,ab,kf.)
557. (((graft vs host or graft versus host) adj (disease$ or syndrome$ or disorder)) and chronic$).ti,ab,kf.
558. exp HIV/
559. exp HIV Infections/
560. (HIV or human immunodeficiency virus$).ti,ab,kf.
561. (htlv or human t-lymphotropic virus$ or human t cell lymphotropic virus$).ti,ab,kf.
562. (acquired immune deficiency syndrome$ or acquired immunodeficiency syndrome$).ti,ab,kf.
563. (AIDS adj3 (virus$ or infection$)).ti,ab,kf.
564. (AIDS adj (related or associated)).ti,ab,kf.
565. exp Neoplasms/
566. (cancer$ or carcin$ or tumor$ or tumour$ or neoplas$ or adenocarcin$ or oncol$ or malignan$).ti,ab,kf.
567. Cystic Fibrosis/
568. (cystic fibrosis or fibrocystic or fibro-cystic or mucoviscidosis or cf).ti,ab,kf.
569. Cerebral Palsy/

570. (cerebr$ adj3 pals$).ti,ab,kf.
571. Muscle Spasticity/
572. spasticit$.ti,ab,kf.
573. Quadriplegia/
574. (spastic$ and (quadripleg$ or tetrapleg$)).ti,ab,kf.
575. exp Renal Insufficiency/
576. ((kidney$ or renal) adj3 (failure$ or insufficienc$)).ti,ab,kf.
577. (end stage adj3 (kidney or renal)).ti,ab,kf.
578. (("stage 5" or "stage V") adj3 (kidney or renal)).ti,ab,kf.
579. (ESRD or ESKD or ESRF or ESKF or CRF or CKF).ti,ab,kf.
580. or/22-579
581. Qualitative Research/
582. Interview/
583. Personal Narratives/
584. Grounded Theory/
585. Focus Groups/
586. Hermeneutics/
587. Anthropology, Cultural/
588. qualitative.af.
589. interview$.af.
590. findings.ti,ab.
591. experiences.ti,ab.
592. views.ti,ab.
593. perspective$.ti,ab.
594. beliefs.ti,ab.
595. attitude$.ti,ab.
596. narrative.ti,ab.
597. ethnograph$.ti,ab.
598. (case study or case studies).ti,ab.
599. thematic analysis.ti,ab.
600. themes.ti,ab.
601. grounded theory.ti,ab.
602. field notes.ti,ab.
603. audio record$.ti,ab.
604. focus group$.ti,ab.
605. conversation$ analys$.ti,ab.
606. descriptive stud$.ti,ab.
607. discourse analys$.ti,ab.
608. exploratory analys$.ti,ab.
609. exploratory stud$.ti,ab.
610. Hermeneutic$.ti,ab.
611. naturalistic.ti,ab.
612. phenomenolog$.ti,ab.

613. participatory.ti,ab.
614. semi structured.ti,ab.
615. key informant$.ti,ab.
616. cultural anthropology.ti,ab.
617. narration.ti,ab.
618. narrative analysis.ti,ab.
619. inductive.af.
620. content analysis.ti,ab.
621. discourse analysis.ti,ab.
622. or/581-621
623. 8 and 21 and 580 and 622

Appendix 3: Quality Appraisal Table for Included Studies

Table a; quality appraisal of included studies using modified CASP tool

		Cancer															CHD
Question	Study	Brody, 2007	Chamberlain, 2007	Chesler, 2001	Clarke, 2005	Cluley, 2015	Hill, 2009	Jones, 2003	Mojica, 2016	Neil-Urban, 2002	Nicholas, 2009	Ogg, 1997	Robinson, 2019	Wolff, 2010	Wolff, 2011	Wills, 2009	Bright, 2016
	Rating	Medium	Medium	Medium	Low	High	High	Medium	Low	Medium	High	Medium	Medium	High	High	Medium	Low
1. Was there a clear statement of the aims of the research?		Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
2. Is the qualitative methodology appropriate?		Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
3. Was the research design appropriate to address the aims of the research?		Yes	Yes	Somewhat	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
4. Are the studies theoretical underpinnings clear, consistent and conceptually coherent?		No	No	No	No	Yes	Yes	No	Can't tell	No	Yes	Yes	No	Somewhat	Somewhat	No	No
5. Was the recruitment strategy appropriate to the aims of the research?		Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
6. Was the data collected in a way that addressed the research issue?		Yes	Yes	Somewhat	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Somewhat	Yes	Yes	Somewhat	Somewhat
7. Has the relationship between researcher		Can't tell	Can't tell	Somewhat	Can't tell	Yes	Yes	Can't tell	Can't tell	Can't tell	Yes	Can't tell	Somewhat	Yes	Yes	Can't tell	No

Question		Cancer															CHD
	Study	Brody, 2007	Chamberlain, 2007	Chesler, 2001	Clarke, 2005	Cluley, 2015	Hill, 2009	Jones, 2003	Mojica, 2016	Neil-Urban, 2002	Nicholas, 2009	Ogg, 1997	Robinson, 2019	Wolff, 2010	Wolff, 2011	Wills, 2009	Bright, 2016
	Rating	Medium	Medium	Medium	Low	High	High	Medium	Low	Medium	High	Medium	Medium	High	High	Medium	Low
and participants been adequately considered?																	
8. Have ethical issues been taken into consideration?		Yes	Yes	Yes	Can’t tell	Yes	Yes	Can’t tell	Yes	Can’t tell	Yes	Yes	Somewhat	Can’t tell	Can’t tell	Somewhat	Can’t tell
9. Was the data analysis sufficiently rigorous?		Yes	Yes	Yes	Somewhat	Yes	Yes	Yes	Somewhat	Yes	Yes	Yes	Somewhat	Yes	Yes	Somewhat	Can’t tell
10. Is there a clear statement of findings?		Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
11. How valuable is the research?		a, b, c*	a, b, c*	a, b, c*	a, c*	a, b, c*	a, b, c*	a, b, c*	a, b, c*	b*	a, b, c*	a, b, c*	a, b , c*	a, b, c*	a, b, c*	a, b, c*	a, b, c*

*a. Findings considered in relation to existing research
b. Discussion relating to implications of findings upon practice or policy
c. Identification of areas in which further research is necessary

Table b; quality appraisal of included studies using modified CASP tool continued

Question		Congenital Heart Defect				Cystic Fibrosis			Genetic conditions	Life-limiting conditions						Neurological conditions	
	Study	Bruce, 2016	Clark, 1999	Gower, 2016	Robinson, 2019	Hayes, 2008	Priddis, 2010	Shardonofsky, 2009	Rivard, 2014	Bailey-Pearce, 2017	Davies, 2013	Davies, 2004	Nicholas, 2016	Rigby, 2012	Ware, 2007	Applebaum, 2012	Lucca, 2016
	Rating	High	Medium	High	Low/Medium	High	Low	Medium	High	High	High	High	High	Medium	High	High	Low/Medium
1. Was there a clear statement of the aims of the research?		Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes

		Congenital Heart Defect				Cystic Fibrosis			Genetic conditions	Life-limiting conditions						Neurological conditions	
Question	Study	Bruce, 2016	Clark, 1999	Gower, 2016	Robinson, 2019	Hayes, 2008	Priddis, 2010	Shardonofsky, 2009	Rivard, 2014	Bailey-Pearce, 2017	Davies, 2013	Davies, 2004	Nicholas, 2016	Rigby, 2012	Ware, 2007	Applebaum, 2012	Lucca, 2016
	Rating	High	Medium	High	Low/Medium	High	Low	Medium	High	High	High	High	High	Medium	High	High	Low/Medium
2. Is the qualitative methodology appropriate?		Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
3. Was the research design appropriate to address the aims of the research?		Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
4. Are the studies theoretical underpinnings clear, consistent and conceptually coherent?		Yes	No	Somewhat	Can’t tell	No	No	Yes	Yes	Yes	Somewhat	Yes	Somewhat	Somewhat	Yes	Yes	No
5. Was the recruitment strategy appropriate to the aims of the research?		Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
6. Was the data collected in a way that addressed the research issue?		Yes	Yes	Yes	Somewhat	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
7. Has the relationship between researcher and participants been adequately considered?		Yes	Can’t tell	Somewhat	Can’t tell	Can’t tell	Somewhat	Yes	Can’t tell	Yes	Can’t tell	Somewhat	Somewhat	Somewhat	Yes	Yes	Can’t tell
8. Have ethical issues been taken into consideration?		Yes	Can’t tell	Yes	Can’t tell	Yes	Somewhat	Somewhat	Some- what	Yes	Somewhat	Yes	Yes	Somewhat	Yes	Can’t tell	Can’t tell
9. Was the data analysis sufficiently rigorous?		Yes	Somewhat	Yes	Yes	Yes	Yes	Somewhat	Yes	Yes	Yes	Yes	Yes	Somewhat	Yes	Yes	Some-what

Congenital Heart Defect						Cystic Fibrosis			Genetic conditions	Life-limiting conditions						Neurological conditions	
Question	Study	Bruce, 2016	Clark, 1999	Gower, 2016	Robinson, 2019	Hayes, 2008	Priddis, 2010	Shardo-nofsky, 2009	Rivard, 2014	Bailey-Pearce, 2017	Davies, 2013	Davies, 2004	Nicholas, 2016	Rigby, 2012	Ware, 2007	Applebaum, 2012	Lucca, 2016
	Rating	High	Medium	High	Low/Medium	High	Low	Medium	High	High	High	High	High	Medium	High	High	Low/Medium
10. Is there a clear statement of findings?		Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
11. How valuable is the research?		a,b*	a, b, c*	a, b, c*	a, b*	a, b, c*	a, b, c*	a, b*	a, b, c*	a, b, c*	a, b c*	a, b, c*	a, b, c*	a, b, c*	a, b, c*	a, b, c	a, b, c*

*a. Findings considered in relation to existing research
b. Discussion relating to implications of findings upon practice or policy
c. Identification of areas in which further research is necessary

Appendix 4: 'The health of mothers of children with a life-limiting condition: a qualitative interview study' (Palliative Medicine publication)

PALLIATIVE MEDICINE

Original Article

Palliative Medicine
2022, Vol. 36(9) 1418–1425

Article reuse guidelines:
sagepub.com/journals-permissions
DOI: 10.1177/02692163221122325
journals.sagepub.com/home/pmj
SAGE

The health of mothers of children with a life-limiting condition: A qualitative interview study

Victoria Fisher[1], Karl Atkin[2] and Lorna K Fraser[1]

Abstract

Background: The number of children with a life-limiting condition is increasing. The mothers of these children commonly provide extensive care at home for their child and are at a higher risk of poor health than other mothers. The impact of this is rarely explored from mothers' perspectives.

Aim: To explore mothers' accounts of their physical and mental health, e xperiences of accessing healthcare and who they think should support their health.

Design: Qualitative semi-structured interviews were conducted and analysed using thematic analysis.

Setting/participants: Thirty mothers of children with a life-limiting condition were recruited via three UK children's hospices and social media.

Results: Mothers felt that their health concerns could be misunderstood by professionals, describing untimely and inappropriate support that failed to recognise the nature of caring for a child with a life-limiting condition. This led to mothers' reluctance in addressing these concerns. Mothers felt unable to prioritise their own needs, relative to those of their child and worried about who would look after their child if they did become unwell. They described stress as a result of battles with services rather than as a result of caregiving. Mothers valued feeling recognised as caregivers, which made it easier to look after their health alongside their child's. Hospice support was particularly valuable in this respect.

Conclusions: A more unified system that recognises not only the unique set of challenges presented to mothers caring for a child with a life-limiting condition, but the value of palliative care services in supporting these mothers, is required.

Keywords

Life-limiting condition, child, paediatrics, palliative care, qualitative

What is already known about the topic?

- Mothers of children with a life limiting condition are more likely to experience physical and psychological health problems than other mothers.
- Understanding the health of these mothers is important in its own right, as well as in ensuring that they feel able to care for their child.

What this paper adds?

- Mothers' health and wellbeing was closely linked to how well they felt their child's needs were being addressed.
- Health and wellbeing problems were more often linked to `battles' with service provision, rather than as a direct result of caregiving.
- Mothers' felt that their unique experiences as caregivers to children with life-limiting conditions could be overlooked, leading to the offer of untimely and inappropriate support and treatment.

---

[1] Martin House Research Centre, Department of Health Sciences, University of York, York, UK
[2] Department of Sociology, University of York, York, UK

Corresponding author:
Victoria Fisher, Martin House Research Centre, Department of Health Sciences, University of York, Area 2 Seebohm Rowntree Building, Heslington, York YO10 5DD, UK.
Email: victoria.fisher@york.ac.uk

Appendix 5: Topic guide for maternal health study

Research study: Crisis Prevention rather than Crisis Management; the Health of Mothers of Children with a Life-Limiting condition

Topic guide for interviews with mothers

1. Introducing the interview and consenting process
    - Researcher introduces self
    - Explain/reiterate: confidentiality, length of interview, nature of discussion, withdraw at any time, fine to take breaks
    - Any questions
    - Obtain consent
    - Start recording

2. Finding out about the child and family
    - Exemplar opening scripts: "To start off, please could you tell me a little bit about [name of child]"; "… and the rest of your family?..."

    - Probes:
        - child's favourite activities; nursery/school; personality
        - siblings and ages; family composition
        - name of health condition
        - history of diagnosis

3. Their own health
    - Exemplar opening script: "And now, please could you tell me a bit about your own health?..."

    - Probes:

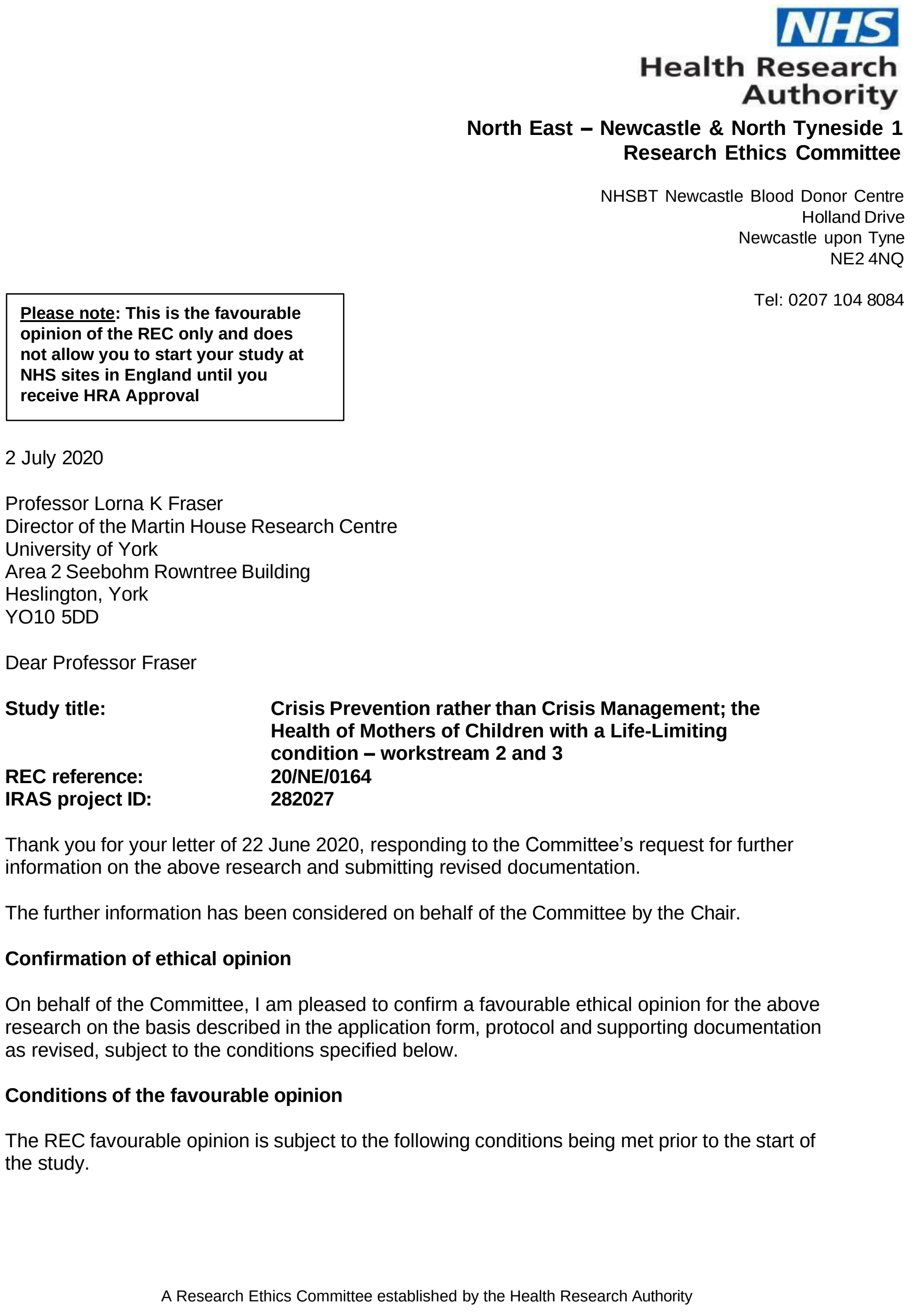
    - Concerns over their psychological health
    - Any physical health conditions
    - Use of medications
    - Factor they feel affect their health both protective and not, e.g. social support, family
    - When did you last see your GP about your health?

4. Follow-up questions on target topic areas:
    - Views on how easy it is to access healthcare for themselves
    - Views on how health has changed over time and in relation to their child's illness
    - Views on which services should help them look after their own health

    - Views on how these services could do that e.g. flexible options, remote consultations, opening hours etc

5. Close:
    - Express thanks and note moving towards end of interview
    - Ask if anything else they would like to add
    - Remind re confidentiality, next steps in research and when findings will be ready
    - Check if any questions
    - Arrange follow-up contact
    - Reiterate appreciation for time and sharing their story.

Appendix 6: REC approvals for maternal health study



NHS
Health Research
Authority
North East – Newcastle & North Tyneside 1
Research Ethics Committee

NHSBT Newcastle Blood Donor Centre
Holland Drive
Newcastle upon Tyne
NE2 4NQ

Tel: 0207 104 8084

Please note: This is the favourable opinion of the REC only and does not allow you to start your study at NHS sites in England until you receive HRA Approval

2 July 2020

Professor Lorna K Fraser
Director of the Martin House Research Centre
University of York
Area 2 Seebohm Rowntree Building
Heslington, York
YO10 5DD

Dear Professor Fraser

Study title:	**Crisis Prevention rather than Crisis Management; the Health of Mothers of Children with a Life-Limiting condition – workstream 2 and 3**
REC reference:	**20/NE/0164**
IRAS project ID:	**282027**

Thank you for your letter of 22 June 2020, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Conditions of the favourable opinion

The REC favourable opinion is subject to the following conditions being met prior to the start of the study.

A Research Ethics Committee established by the Health Research Authority

Appendix 7: Quantitative survey

The health of fathers of children with a life-limiting condition- Qualtrics survey

[Fathers referred by healthcare professionals will have received a hard copy of the information sheet and will be directed straight to consent page- with the option of reading information sheet again (downloadable pdf.)].

You are invited to take part in a research study about the health of fathers of children with a life-limiting condition being carried out by researchers at The Martin House Research Centre at the University of York (https://www.york.ac.uk/healthsciences/research/public-health/projects/martinhouse/). This includes all individuals that define themselves as the fathers of a child with a life-limiting condition including biological fathers, non-biological fathers, foster-fathers, step-fathers and adoptive fathers. Before you decide to take part, it is important that you understand the nature of the study. Please read the following details carefully. If you have any questions before you consent to take part, please feel free to contact the research team (victoria.fisher@york.ac.uk, 01904 321641). If you would like to complete the survey over the phone, then this can be arranged by contacting the research team.

Why is this research being carried out?
There are more children and young people living with medical conditions that may shorten their lives (life-limiting conditions). The parents of these children commonly provide and coordinate all of their child's care needs. The existing research on these parents mainly focuses on mothers, meaning that we know less about fathers. **This research study is important because it will help us to understand the experiences of fathers and how their experiences might affect their own health.** It will also help us to understand how fathers look after their own health and well-being. Understanding the health of fathers is important for fathers themselves but also to make sure that they feel able to care for their child.

Why have I been invited to take part?
You are invited to take part if you are the father of a child who has been diagnosed with a life-limiting or life-threatening condition that may shorten their life. You must be aged 18 or over, have a child that is aged 25 or under and has been diagnosed with a life-limiting condition and be fluent in English to participate.

Eligibility:

1. Are you a father of a child who has a life-limiting condition?

a. Yes

b. No (if no, cannot continue and will be taken to ineligibility statement)

2. Please enter your date of birth: <drop down boxes for month and year>

3. Please enter your child's date of birth: <drop down boxes for month and year>

Survey ineligibility:

Thank you very much for your interest in our survey. However, you are not eligible to take part. Please feel free to contact the research team if you have any questions:

Victoria.fisher@york.ac.uk, 01904 431641

You can now close the page to leave the survey.

If eligible:

What will happen if I decide to take part?

You will be asked to answer some questions about you, your family, your child's characteristics including their health condition and then some questions about your physical and mental health, sleep and caregiving. The survey will take around 20 minutes to complete, and we ask that you complete it as accurately as possible. Your participation is voluntary and all of the answers that you give will be kept confidential. At the end of the survey, you may also provide us with your name and contact information if you are interested in taking part in an interview with a researcher to help us to understand your experiences in more detail.

Do I have to take part?

No- the study is completely voluntary. If you do decide to take part, you can withdraw at any point by closing your internet browser. Deciding not to take part or to withdraw will not affect you, or any care or benefits that you or your child receive.

Will my taking part be confidential?

All of the information we collect during the course of the research will be kept confidential and there are strict laws to safeguard your privacy at every stage. You can find out more about how we use your information at https://www.york.ac.uk/records-management/dp/ or by contacting dataprotection@york.ac.uk.

Can I change my mind?

You can withdraw at anytime during the survey by closing your internet browser. If you choose to withdraw once you have submitted your responses, you can do so (by contacting victoria.fisher@york.ac.uk, 01904 321641) as long as you have provided your contact details at the end of the survey. Your responses will **not be** included in the study as long as they have not already been used for analysis. If you do not provide your contact details, then once your responses are submitted they cannot be removed. This is because we will have no way of knowing which were your responses.

What are the benefits of taking part?

By taking part in this study, you could help others. Information collected from you and from other fathers could lead to a better understanding of the experiences and health of fathers of children with a life-limiting condition.

Are there any risks to taking part?

It is possible that you may find it difficult or upsetting to answer questions relating to your own health. Taking part is voluntary and you can decide to withdraw from the study at any time.

What will you do with the information I provide?

We will use the information you provide in this survey to describe the health of fathers of children with a life-limiting condition, and factors that influence this. Individuals from the University of York and regulatory organisations may look at your research records to check the accuracy of the research study. The only people at the University of York who will have access to information that identifies you will be the people that need to contact you or audit the data collection process. The people who analyse the data will not be able to identify you. If you choose not to provide your contact details, all of your data will remain anonymous. We will write about the study in publications read by researchers and care providers. We may present the findings at events and on websites.

The information that we share with others will not identify you and will only be used for the purposes of health and social care research. It cannot be used to contact you and will not affect the care that you or your child receive. It will not be used to make decisions about future services available to you, such as insurance.

All electronic survey data will be stored on a password-protected folder on university computers. This will be stored securely for at least 10 years.

Who can I contact?

If you have any questions about the study please contact: victoria.fisher@york.ac.uk, 01904 321641.

Consent Statements (all mandatory)

I have read and understood the study information <tick box>

I understand what the study is about, what taking part involves and that taking part is voluntary <tick box>

I know that any information I provide will be stored securely at the University of York <tick box>

I know that the findings from this project will be shared with the public and on websites and that this will **not** include my name or personal details <tick box>

I agree to take part in the study <tick box>

<Click here to start>

Survey Questions:

SECTION 1: About you

1a. In what region do you currently reside

- East of England
- East Midlands
- London
- North East
- North West
- Northern Ireland
- Scotland
- South East
- South West
- Wales
- West Midlands
- Yorkshire and the Humber

- Other

2a. Were you born in the UK?
- Yes
- No

If no 2b: What country were you born in?
  2c: In what year did you move to the UK? <please type yyyy>

3. Which of the following best describes the area in which you live?
- Large city
- Small city or large town
- Suburban area
- Village or rural area

4. What is your ethnic group?

White
- English/Welsh/Scottish/Northern Irish/ British
- Irish
- Gypsy or Irish Traveller
- Any other white background, please describe <free text box>

Mixed/ Multiple ethnic groups
- White and Black Caribbean
- White and Black African
- White and Asian
- Any other Mixed/ Multiple ethnic background, please describe <free text box>

Asian/ Asian British
- Indian
- Pakistani
- Bangladeshi
- Chinese
- Any other Asian background, please describe

Black/ African/ Caribbean/ Black British
- African
- Caribbean
- Any other Black/ African/ Caribbean background, please describe <free text box>

Other ethnic group
- Arab
- Any other ethnic group, please describe <free text box>

5. What is your religion?
- Christian (including Church of England, Catholic, Protestant and all other Christian denominations)
- Buddhist
- Hindu
- Jewish
- Muslim
- Sikh
- No religion
- Other religion, please describe <free text box>

6. Which of the following, if any, is the highest educational or professional qualification you have obtained?
- GCSE/O-Level/CSE
- Vocational qualifications (NVQ1/NVQ2)
- A-Level/Scottish Higher or equivalent (NVQ3)
- Bachelor’s degree or equivalent (NVQ4)
- Masters/PhD or equivalent
- No formal qualifications
- Other

7. Which of the following best describes your current occupation?
- In full-time work
- Part-time or casual work
- Unemployed but looking for work
- Unemployed and not looking for work
- Home/ caring duties
- Retired
- Student
- Other <please specify>

8. Which of the following income categories best describes your total household income per year before taxes?
- Less than £10,000
- £10,000 to £24,999

- £25,000 to £49,999
- £50,000 to £74,999
- £75,000 to £99,999
- £100,000 or more

9. Relationship status?
- Single; never married
- Married or domestic partnership
- Divorced
- Separated
- Widowed

10. Which of the following best describes your household?
- Living alone
- A lone parent household with child/ren (including child with a life-limiting condition)
- A lone parent household with child/ren but child with life-limiting condition does not live with you
- Living with partner/ spouse and child/ren (including child with a life-limiting condition)
- Living with partner/spouse and child/ren but child with life-limiting condition does not live with you
- Other (please specify)

11. How many children do you have? <add number to box>

12. Please enter the age and sex of each of your children and identify which child/ren have a life-limiting condition <drop down boxes for age and sex, yes/no for life-limiting condition>

SECTION 2: Your child

If you have more than one child, these questions relate to the child diagnosed with a life-limiting condition. If you have more than one child diagnosed with a life-limiting condition, you will be able to provide additional details once you have entered details for the first child.

What is your child's main diagnosis? <free text box>

13. At what age was your child diagnosed with this condition? <select age from drop down box, including N/A for undiagnosed children>

14. How would you best describe your child's care needs relative to other children of a similar age?

- Significantly more care; most or all of the time
- Moderately more care most of the time
- Slightly more care most of the time
- The same care

15. Has your employment status changed since your child was diagnosed?
- Stopped work to care for child
- Reduced work hours to care for child
- Change in career path
- No change in employment
- Changed for other reason

16. How would you describe your caregiving role?
- Child's primary caregiver
- Joint caregiving responsibilities with child's other parent
- Your child's other parent is their primary caregiver
- Other <please specify>

17. Does your child/ family receive support from a hospice?
- Yes
- No

SECTION 3: Your own health

18. How is your health in general?
- Very good
- Good
- Average
- Poor
- Very poor

19a. Do you have any physical or mental health conditions or illnesses lasting or expected to last 12 months or more?
- Yes
- No

If yes 19b. What is the nature of your health condition/s? <tick box>
- Arthritis
- A speech impairment
- Chest or breathing problems (asthma/bronchitis)

- Diabetes
- Difficulty hearing
- Difficulty seeing (even when wearing spectacles/ contact lenses)
- Dyslexia
- Epilepsy
- Heart, blood pressure or circulation problems
- Learning or behavioural problems e.g. autism
- Mental health problems e.g. anxiety or depression
- Problems or disabilities related to arms or hands
- Problems or disabilities related to legs or feet
- Problems or disabilities related to back or neck
- Severe disfigurement, skin condition or allergies
- Severe stomach, liver, kidney or digestive problems
- Some other progressive disability or illness
- Difficulty understanding spoken and/or written word
- Some other health problem or disability <please specify>

20. Please respond to each question by marking one box per row

In the past 7 days…
- My sleep quality was <very poor, poor, fair, good, very good>

In the past 7 days…
- My sleep was refreshing <not at all, a little bit, somewhat, quite a bit, very much>
- I had a problem with my sleep <not at all, a little bit, somewhat, quite a bit, very much>
- I had difficulty falling asleep <not at all, a little bit, somewhat, quite a bit, very much>
- My sleep was restless <not at all, a little bit, somewhat, quite a bit, very much>
- I tried hard to get to sleep <not at all, a little bit, somewhat, quite a bit, very much>
- I worried about not being able to fall asleep <not at all, a little bit, somewhat, quite a bit, very much>
- I was satisfied with my sleep <not at all, a little bit, somewhat, quite a bit, very much>

21. Under each heading, please tick ONE box that best describes your health TODAY

22. Mobility

- I have no problems in walking about
- I have slight problems in walking about
- I have moderate problems in walking about
- I have severe problems in walking about
- I am unable to walk about

23. Self-care

- I have no problems washing or dressing myself
- I have slight problems washing or dressing myself
- I have moderate problems washing or dressing myself
- I have severe problems washing or dressing myself
- I am unable to wash or dress myself

24. Usual activities (e.g. work, study, housework, family or leisure activities)

- I have no problems doing my usual activities
- I have slight problems doing my usual activities
- I have moderate problems doing my usual activities
- I have severe problems doing my usual activities
- I am unable to do my usual activities

25. Pain/ Discomfort

- I have no pain or discomfort
- I have slight pain or discomfort
- I have moderate pain or discomfort
- I have severe pain or discomfort
- I have extreme pain or discomfort

26. Anxiety/ Depression

- I am not anxious or depressed
- I am slightly anxious or depressed
- I am moderately anxious or depressed
- I am severely anxious or depressed
- I am extremely anxious or depressed

27. Visual analogue scale (0-100)

- We would like to know how good or bad your health is TODAY.
- This scale is numbered from 0 to 100.
- 100 means the <u>best</u> health you can imagine.
- 0 means the <u>worst</u> health you can imagine.
- Please mark an X on the scale to indicate how your health is TODAY.
- Now, write the number you marked on the scale in the box below.

Section 4: Your caregiving experiences

28. Please rate each item below between 5= strongly agree and 1= strongly disagree

29. Caregiver strain

- As a caregiver, I feel tired and run down
- As a caregiver, I feel my own health has suffered
- As a caregiver, I feel I am losing control over my own life
- As a caregiver, I don't have enough time for myself
- I feel isolated and alone in caring for my child
- I have had to give up my social life to care for my child
- As a caregiver, I gave not been able to do my job or study as well as I would like
- Caring for my child creates financial difficulties

30. Positive appraisal of caregiving

- Caring for my child is satisfying
- It is a privilege to care for my child
- Caring for my child has made me feel close to him/her
- I am able to comfort my child when he/she needs it
- I feel confident I can handle most problems when caring for my child
- I feel useful in my relationship with my child
- I am committed to caring for my child

31. Caregiver distress

- I feel guilty about not being able to do more for my child
- I worry that I won't be able to do enough to care for my child
- I feel anxious about caring for my child
- I feel depressed about caring for my child

32. Family well-being

- Our family works together to solve problems
- Our family is able to talk about our feelings with each other
- I feel our family is closer because of caring for our child
- Because of caring for my child our family is better able to cope with change
- Our family disagrees a lot caring for my child
- Our family avoids discussing their fears and concerns about caring for my child

The next part of the project is an optional interview with a researcher.

If you are interested in taking part, or would like some more information before you decide, please provide your contact information below and a researcher will be in touch to discuss it with you and to organise an interview if appropriate. The aim of the interviews is to explore fathers' experiences in more detail. If you do not want to take part in an interview, you can now exit the survey. *Thank you very much for taking the time to share your responses with us.*

Contact details to discuss interview:

Name: <free text box>
Email address: <free text box>
Telephone number: <free text box>
Preferred means of contact Email (tick box) Telephone (tick box)
I give consent for a researcher to contact me about the next part of the study (tick box)

<submit>

Appendix 8: Invitation letter for fathers

<Site name and address>

Re: Invitation to take part in research about fathers

Dear x,

We would like to invite you to take part in a research study looking at the health of fathers of children with a life-limiting condition. This is being carried out by researchers at the University of York. It is important because much of the research in this area has been with mothers, meaning that we know less about the experiences and health of fathers. The aim of the study is to learn more about the health of fathers, things that affect their health and how this information can be used to identify appropriate support for fathers.

The study is made up of 2 parts; an online survey and an interview. You can take part in just the survey or both parts, depending on your preference. Included with this letter is a participant information sheet which gives you more information about the study and what taking part involves. If you decide to take part, the link to the internet survey is below. It is also on the information sheet. If you have any questions, please do not hesitate to contact the research team (victoria.fisher@york.ac.uk).

[ADD link to survey]

Best wishes,

<clinician and site name>

Appendix 9: Social media advert

Are you a father of a child with a life-limiting condition?

We would like to invite you to take part in a research study looking at the health of fathers of children with a life-limiting condition. Much of the research in this area has been with mothers, meaning that we know less about the experiences and health of fathers.

There are 2 parts to the study; **Part A** is a web-based survey that you can complete in your own time.

Part B is an interview with a researcher that will help us to get more of an in-depth understanding of your experiences.

You can take part in *Part A only* or *both parts* of the study

Please follow the link to the survey or scan the QR code below:

If you decide to take part in part B as well, you will be able to provide your contact details at the end of the survey and a researcher will contact you to arrange the interview.

If you would like to complete the survey over the phone, please contact Victoria Fisher victoria.fisher@york.ac.uk or 01904 321641 and this can be arranged

Appendix 10: Information sheet for fathers

Research study

The health of fathers of children with a life-limiting condition Information for fathers

- We would like to invite you to take part in a research study to learn more about the health of fathers of children with a life-limiting condition. This includes all individuals who define themselves as the father or a child with a life-limiting condition including biological fathers, non-biological fathers, step-fathers, foster fathers and adoptive fathers.
- The study will collect information from fathers of children with a life-limiting condition and is being carried out by researchers at The Martin House Research Centre at the University of York (https://www.york.ac.uk/healthsciences/research/public-health/projects/martinhouse/)

Why is this research being carried out?

There are more children and young people living with medical conditions that may shorten their lives (life-limiting conditions). The parents of these children commonly provide and coordinate all of their child’s care needs. The existing research on these parents focuses on mothers, meaning that we know less about fathers. This research study is important because it will help us to understand the experiences of fathers and how their experiences might affect their own health. It will also help us to understand how fathers look after their own health and well-being. Understanding the impact on fathers' own health is important not only for fathers themselves but in ensuring that they feel able to care for their child.

Why have I been chosen, and do I have to take part?

You have been identified as the father of a child with a life-limiting condition. Taking part is entirely your decision and whatever you decide will **not** affect the standard of care and support or other benefits you and your child receive.

What will happen if I decide to take part?

The study is made up of 2 parts: a web-based survey and an interview with a researcher. If you take part in the study you will be asked to complete the survey first. You will then be asked if you would also like to take part in an interview. This is optional though.

The Survey

The survey is web-based and you can do it in your own time. It will take around 20 minutes to complete. You will be provided with a web link to the survey. There will be a short section at the beginning to check that you are eligible and then you will be able to consent to take part. The main part of the survey will include questions relating to you, your family, your child's characteristics including their health condition, and your own physical and mental health, sleep and caregiving. If you decide that you are interested in taking part in the interview as well as the survey, you will be able to provide your contact details at the end of the survey.

The interview

This involves an interview with a researcher which we will use to gain more of an in-depth understanding of your experiences and health. This interview will last around 45-90 minutes. Once we have your details, we will contact you to arrange an interview and answer any questions that you may have. During the interview you will be asked to talk about your experiences as a father of a child with a life-limiting condition, your own physical and mental health and how you think your experiences have affected your health. You will also be asked about ways in which you look after your own health.

You can choose where the interview takes place and you can have someone with you if you would like. It can be face-to-face (depending on COVID-19 restrictions), over the phone or via video call e.g. Zoom. With your consent, the interview will be audio recorded so that we don’t miss anything that you tell us.

Will my taking part be kept confidential?

We will keep your survey responses and what you tell us in interview or during contact with the research team completely confidential, unless you tell us something that raises concerns that you or someone else is at serious risk of harm. If this happens, we would talk to you first and the best thing to do. We will follow strict rules about confidentiality and all information will be stored in locked filing cabinets and in password-protected folders on computers.

What are the possible benefits and risks to me of taking part?

By taking part in this study you could help others. Information collected from you and from other fathers could lead to a better understanding of the experiences and health of fathers of children with a life-limiting condition. It is possible that you may find it difficult to talk about or answer questions relating to your own health. If you find the survey or the interview distressing, please contact the research team and we can provide details of local organisations who can help. Taking part is voluntary and you can decide to withdraw from the study at any time.

Are there any expenses or payment for taking part in this study?

If you have to travel in order to participate in the study, we will reimburse your travel costs.

If I want to take part, what do I need to do next?

If you would like to take part in the survey, please follow the URL link below or scan the QR code where you can consent to the study and complete the survey.

https://york.qualtrics.com/jfe/form/SV_7NVsrVKvBOXoTEq

If you would also like to take part in the interview, then you will be able to provide your contact details at the end of the survey and a researcher will contact you. Whether you take part in the survey, or both parts is completely your choice.

If you have any questions about either aspect of the study or about any of the statements on this form or the consent form please contact Victoria Fisher, victoria.fisher@york.ac.uk, 01904 321641.

What if I change my mind during the study?

You can withdraw at any time and you do not need to give us a reason. We will keep the information we have already obtained from you. If we have your personal details we will keep them so that if you contact us again, we know that you have taken part. We will not use this information to contact you though.

What will you do with the information I provide?

We will use the information you provide to describe the health of fathers of children with a life-limiting condition, and factors that influence this. Individuals from the University of York and regulatory organisations may look at your research records to check the accuracy of the research study. The only people at the University of York who will have access to information that identifies you will be the people that need to contact you or audit the data collection process. The people who analyse the data will not be able to identify you. If you choose not to provide your contact details (i.e. only take part in the survey and not the interview), all of your data will remain anonymous. We will write about the study in publication read by researchers and care providers. We will present the findings at events and on websites. We may use the data to support other research in the future, and the data may be shared with other researchers in a way that you will not be identifiable. The information that we share with others will not identify you and will only be used for the purposes of health and social care research. It cannot be used to contact you and will not affect the care that you or your child receive. It will not be used to make decisions about future services available to you, such as insurance.

How long will the University of York keep my information?

The University of York will keep identifiable information about you, including your signed consent form, for 3 years after the study has finished. We will also keep the information you share in the survey and the interview for at least 10 years from the end date of the study. Your interview data will be stored as an anonymised transcript of the audio recording. The audio recording will be deleted immediately after transcription and accuracy checks.

Who is responsible for keeping my information safe?

The University of York is the sponsor this study based in the United Kingdom. We will be using information from you in order to undertake this study and will act as the data controller for this study. This means that we are responsible for looking after your information and using is properly.

The University of York is a publicly-funded organisation that conducts research to improve health, care and services. Research following UK Policy Framework for Health and Social Care Research is conducted to serve the interests of society as a whole. This means that the University of York is using the legal basis provided under the General Data Protection Regulation of a task in the public interest to use your personal data for this research. Your rights to access, change or move your information are limited, as we need to manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the study, we will keep the information that we already obtained about you. To safeguard your rights, we will use the minimum personally identifiable information possible.

You can find out more about how we use your information at https://www.york.ac.uk/healthsciences/research/trials/trials-gdpr/research-partcipants/ or by contacting dataprotection@york.ac.uk.

What will happen if the study stops earlier than expected?

Should the research stop earlier than planned and you are affected in any way, we will tell you and explain why if we have your contact details.

Thank you for the taking the time to read this information sheet. Please keep this copy.

Get in touch with the research team:

Victoria Fisher

e. victoria.fisher@york.ac.uk

mhrc@york.ac.uk

Study funding and approvals:
The University of York is the sponsor for this study.
The study is funded by the National Institute for Health Research (NIHR) (Academy) (Study Reference:18400).

The London- Bloomsbury Research Ethics Committee have approved this study (reference: 21/LO/0591)

Who to contact if you have concerns about the study or wish to make a complaint:
Lorna Fraser- Director of the Martin House Research Centre: email: lorna.fraser@york.ac.uk telephone: 01904321889
Victoria Fisher: email: victoria.fisher@york.ac.uk telephone: 01904321641
If you feel unhappy with the way your complaint was dealt with, please contact the research office at The University of York:
Michael Barber Telephone: 01904 318693
Email: michael.barber@york.ac.uk
Address: Innovation Centre, Ron Cooke Hub, University of York, York, YO10 5GE
If you wish to raise a complaint on how we have handled your personal data, you can contact the University of York's Data Protection Officer who will investigate the matter. If you are not satisfied with our response or believe we are processing your personal data in
a way that is not lawful, you can complain to the Information Commissioner's Office (ICO).

The University of York's Data Protection Officer can be contacted at
dataprotection@york.ac.uk

Appendix 11: Topic guide for fathers' interviews

Research Study: The health of fathers of children with a life-limiting condition; a mixed methods study

1. Introducing the interview and consenting process
- Researcher introduces self
- Explain/ reiterate: confidentiality, length of interview, nature of discussion, explain that participant can withdraw at any time, fine to take breaks
- Does the participant have any questions
- Obtain consent
- Start recording

2. Finding out about the father and their family
- Example opening scripts: "To start off, please could you tell me a little bit about you and your family?..."
- Probes:
  - number of children
  - children's ages and family composition
  - what a typical day is like including experience of caregiving
  - what is your experience of supporting your children?
  - what were your feelings when you first found out there was something wrong with one of your children?

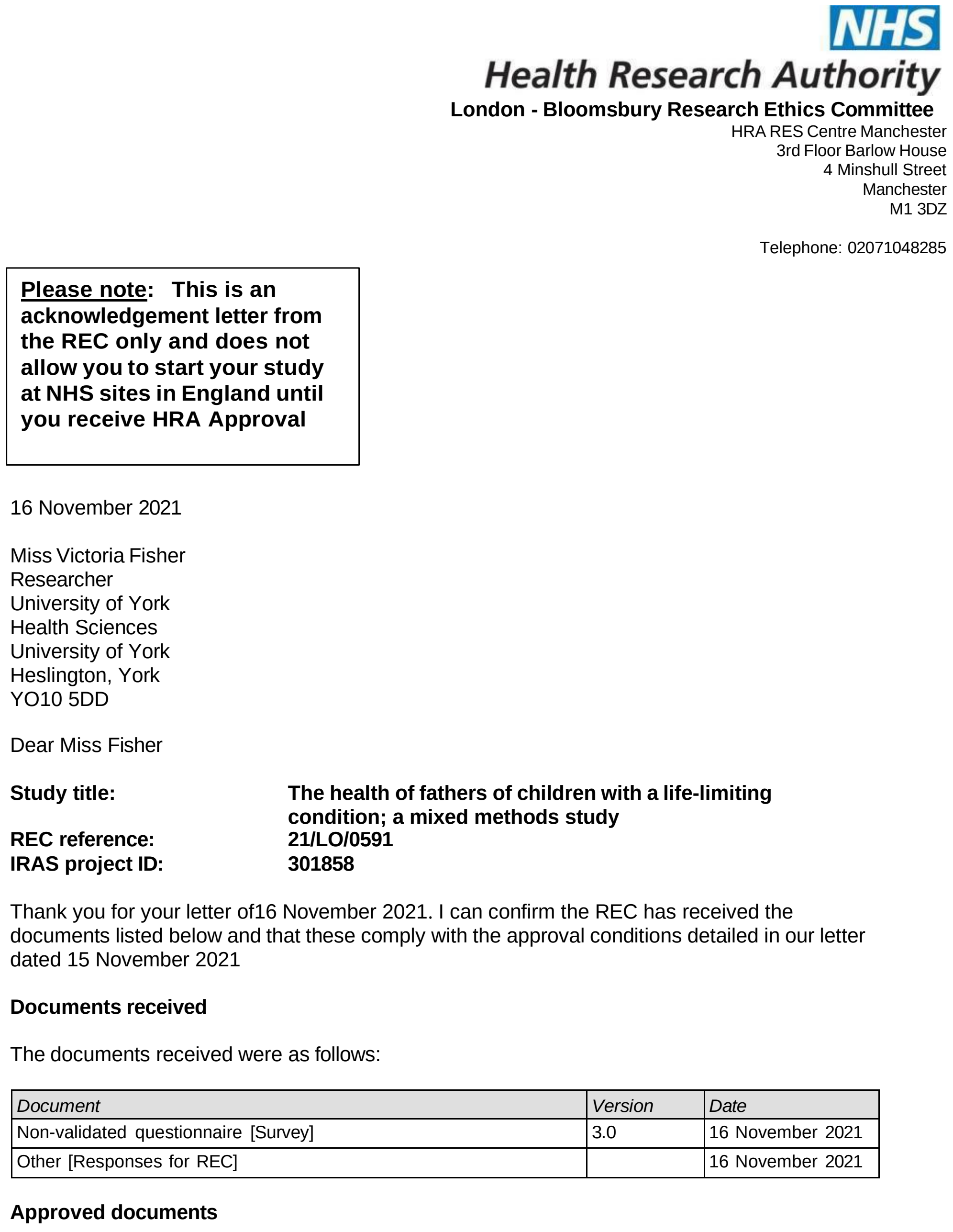
3. Caregiving
- Exploration of fathers' roles as caregivers.
- Probes:
  - Employment
  - Roles in caring for child and other children
  - How/ if role has changed
  - Thoughts/ feeling associated with caregiving/ role

. Fathers' health and wellbeing
- Example opening script: "Now if we could move onto talking a little bit about your own health now, is that okay?"
- Probes:
  - Any concerns with regards to your physical or mental health/ how would you describe your own health?
  - Particular factors that fathers feel may have harmed or have helped/ protected their health
  - Means of support and ease of access; formal or informal
  - Experiences with healthcare professionals
  - When was the last time fathers were asked about their own well-being/ have fathers been made to feel like their own health is important?

5. Follow-up questions on target topic areas:
- Views on how easy it is to access healthcare for themselves
- Workplace support
- Relationships with healthcare staff
- Views on how health has changed over time and in relation to their child's illness
- Coping

6. Close:
- Express thanks and note moving towards end of interview
- Ask if anything else they would like to add
- Remind re confidentiality, next steps in research and when findings will be ready
- Check if any questions
- Arrange follow-up contact
- Reiterate thanks and appreciation for time/ taking part

Appendix 11: REC and HRA approval for fathers' study



NHS
Health Research Authority
London - Bloomsbury Research Ethics Committee
HRA RES Centre Manchester
3rd Floor Barlow House
4 Minshull Street
Manchester
M1 3DZ

Telephone: 02070148285

<u>Please note</u>: This is an acknowledgement letter from the REC only and does not allow you to start your study at NHS sites in England until you receive HRA Approval

16 November 2021

Miss Victoria Fisher
Researcher
University of York
Health Sciences
University of York
Heslington, York
YO10 5DD

Dear Miss Fisher

Study title:	**The health of fathers of children with a life-limiting condition; a mixed methods study**
REC reference:	**21/LO/0591**
IRAS project ID:	**301858**

Thank you for your letter of16 November 2021. I can confirm the REC has received the documents listed below and that these comply with the approval conditions detailed in our letter dated 15 November 2021

Documents received

The documents received were as follows:

Document	*Version*	*Date*
Non-validated questionnaire [Survey]	3.0	16 November 2021
Other [Responses for REC]		16 November 2021

Approved documents

Ymchwil Iechyd
a Gofal Cymru
Health and Care
Research Wales

NHS
Health Research
Authority

Miss Victoria Fisher
Researcher
University of York
Health Sciences
University of York
Heslington, York
YO10 5DD

Email: approvals@hra.nhs.uk
HCRW.approvals@wales.nhs.uk

17 November 2021

Dear Miss Fisher

HRA and Health and Care Research Wales (HCRW) Approval Letter

Study title: **The health of fathers of children with a life-limiting condition; a mixed methods study**
IRAS project ID: **301858**
REC reference: **21/LO/0591**
Sponsor **University of York**

I am pleased to confirm that **HRA and Health and Care Research Wales (HCRW) Approval** has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further relating to this application.

Please now work with participating NHS organisations to confirm capacity and capability, in line with the instructions provided in the "Information to support study set up" section towards the end of this letter.

How should I work with participating NHS/HSC organisations in Northern Ireland and Scotland?
HRA and HCRW Approval does not apply to NHS/HSC organisations within Northern Ireland and Scotland.

If you indicated in your IRAS form that you do have participating organisations in either of these devolved administrations, the final document set and the study wide governance report (including this letter) have been sent to the coordinating centre of each participating nation. The relevant national coordinating function/s will contact you as appropriate.