A qualitative IPA study examining the experience of storytelling among parents of children with life-limiting conditions (LLC) and life-threatening conditions (LTC) in a UK paediatric palliative care context

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Background: A paucity of literature exists concerning parents of children with life-limiting conditions (LLCs) and life-threatening conditions (LTCs). In order to interpret their life world, this thesis includes a qualitative systematic review (SR) and narrative synthesis examining their experiences as caregivers. Storytelling (i.e. the experiential act of telling one's story of caring for a child with LLC / LTC) remains under researched in this population. Hence, this feeds into an empirical research paper, which then investigates experiences of storytelling in parents of children with LLC and LTC.

Method: In-depth semi structured interviews were conducted with 8 parents (6 mothers, 2 fathers) caring for a child with LLC or LTC. Parents were recruited from a UK children's hospice charity and were interviewed regarding their experiences of storytelling. Interviews were subjected to Interpretative Phenomenological Analysis (IPA).

Results: From the IPA five superordinate themes emerged: (a)'bonding with other parents through storytelling' (b) 'therapeutic storytelling to a hospice professional' (c) 'storytelling as an educational tool' (d) 'fear of others reactions to the story' (e) 'weariness through repetition of the story'. Themes suggested parents predominantly benefited from telling their story in a children's hospice context (a, b). Parents also benefited from telling their story publicly in an attempt to educate others (c), although the negative aspects of storytelling predominated in everyday settings in the community (d, e). Supportive and empowering social contexts provided storytelling experiences that were perceived as psychologically positive and therapeutic.

Conclusions: Further research is needed to explore the experience of storytelling in this population. Storytelling has the potential to support meaningful, transformative and cathartic experiences for parents. However, its negative potential impacts also require further examination.

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Chapter 1 is a systematic review and narrative synthesis. It examines parents' experiences of caring for a child with LLC or LTC. Five themes are identified from 12 empirical studies. The results appear to indicate that parents experience positive emotional change and growth, alongside distress when caring for a child with LLC or LTC. Further studies involving this population are advocated.

Chapter 2 connects the results from the systematic review and wider literature (chapter 1) to the empirical research paper (chapter 3). It argues that few studies have explored the factors that directly contribute to positive emotional change in the context of caring for a child with LLC/LTC. In doing so it draws upon Pennebaker's emotional disclosure theory (2000) to consider the therapeutic possibilities that are often associated with the experience of storytelling. A rationale is then provided for further qualitative storytelling research in a paediatric palliative care context involving parents.

Chapter 3 is a qualitative empirical research paper exploring parents' experiences of storytelling both in and outside the context of a children's hospice. Five themes are identified. The paper indicated that parents encounter both positive and negative storytelling experiences when caring for a child with LLC and LTC. Themes reflected the understanding that storytelling is often a psychologically positive and therapeutic experience for parents, particularly within a children's hospice context.

Chapter 4 is an additional methodology chapter. The qualitative IPA methodology employed in chapter 3 is described in further detail.

Chapter 5 is an extended discussion. It seeks to evaluate the findings presented in the empirical paper (chapter 3). The strengths and limitations relating to the thesis are explored. Finally, clinical and research implications are also examined.

1.

Systematic Review

What are the experiences of parents when caring for a child with a life-limiting condition or life-threatening condition? A systematic review and narrative synthesis

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Abstract

Background: Life-limiting and life-threatening conditions in children have significantly increased in prevalence over the last decade. To interpret the needs of parents caring for these children an understanding of their experiences is required.

Objective: This article aimed to systematically review and synthesise published qualitative literature involving parents' experiences of caring for a child with a life-limiting or life-threatening condition.

Method: SCOPUS, PsycINFO, the BNI, Web of Science, CINAHL, AMED and MEDLINE were searched systematically from 1997-2017. Manual searches for further relevant articles were conducted in four additional databases. Relevant articles that met the inclusion criteria were critically analysed and synthesised using the narrative synthesis method of Popay et al (2006).

Results*:* Overall the search yielded 12 eligible articles. Five key themes were identified from the synthesis: (a)'navigating the system'; (b)'burden of care'; (c)'living with uncertainty'; (d)'strength through adversity' and (e)'connecting with other families'.

Conclusion: Themes indicate that parents encounter similar positive and negative experiences across conditions. Parents experience positive emotional change and growth, although they could also benefit from additional nursing and psychological support to manage caregiver burden. Rare illnesses and fathers' experiences require further research. PROSPERO registration no: CRD42017083265.

Keywords:
narrative synthesis, life-limiting condition, life-threatening condition, parent, caring, experience.

Introduction

In recent years paediatric palliative care (PPC) has emerged as a small but distinct subspecialty of medicine (Corkin, Price & Gillespie, 2006). The implementation of PPC is heterogeneous, complex and primarily designed to support critically ill children with life-limiting conditions (LLCs) and life-threatening conditions (LTCs). LLCs and LTCs represent a wide range of illness conditions that require in-depth medical knowledge to precisely diagnose (Patterson, Holm & Gurney, 2004). However, Together for Short Lives™ (TFSL), a UK-based PPC organisation (formerly ACT) has defined four main typologies of LLCs and LTCs throughout childhood (Table 1). According to TFSL (2013) LTCs are conditions that threaten life but may be curable; meanwhile LLCs are defined as conditions where the child's premature death is considered likely or inevitable (Mitchell, Knighting, O'Brien, & Jack, 2016).

Prevalence rates for these illness conditions have increased significantly over the last ten years (Rapoport, Beaune Weingarten, Rugg, Newman, 2012). Recent epidemiological data produced by Fraser et al. (2012) revealed that the overall LLC & LTC prevalence rate in England has increased significantly from 25 per 10,000 in 2000-2001 to 32 per 10,000 in 2009-2010 for children aged 0-19 years. Escalating trends in prevalence by year are presented in figure 1. Approximately 40,000 children are now living with an LLC or LTC in England (Popejoy, Pollock, Almack, Manning, & Johnston, 2017) although this number is rising (Jarvis, Parslow, Carragher, Beresford, Fraser, 2017) and sustainable healthcare practices that can effectively accommodate the needs of this growing clinical population are required (Mitchell, Morris, Bennett, Sajid, & Dale, 2017).

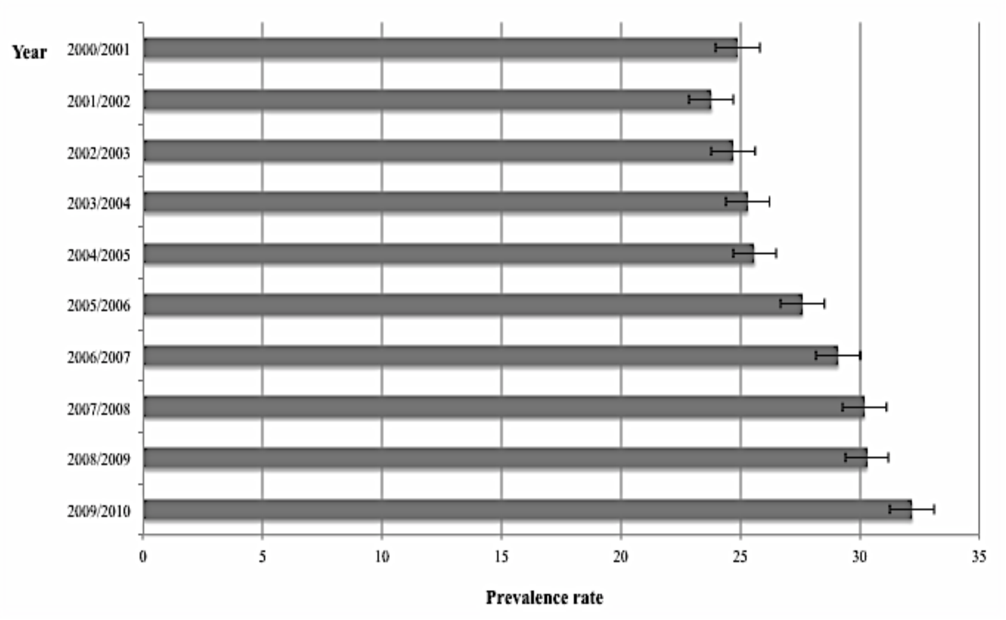

Figure 1. Prevalence of LLCs & LTCs for children in England, 2000-2010, using data obtained from Fraser et al (2012).

In the context of these demographic changes, considerable practical and economic challenges exist that may negatively impact upon the provision of care. Clinical treatment is typically time intensive, costly and resources are limited (Hain, Heckford, & McCulloch, 2012). Due to growing population pressure, as well as ad-hoc funding (Dunbar, 2016), access to hospital based PPC is limited in many areas of the UK (Padget & Cadywould, 2015). Thus, only a small number of children with a LLC or LTC are able to obtain inpatient medical treatment; while the majority of children receive care within the family home via their parents, or other extended family members (Bluebond-Langner, Beecham, Candy, Langner, & Jones, 2013).

An on-going trend towards delivering care inside the home is not without difficulties for parents. Everyday responsibility for managing the child's illness condition within the home falls primarily upon the parent, with community health providers playing only a minor supporting role (Remedios et al., 2015). Parent caregivers play an important role in delivering home centered treatment as they often oversee the administration of medications (Hudson, 2005), develop advanced nursing skills (Wray, Lindsay, Crozier, Andrews, & Leeson, 2013) promote therapeutic interventions (McSherry, Kehoe, Carroll, Kang, & Rourke, 2007) and liaise directly with outside agencies to ensure that care of a highly technical nature is delivered (Rodriguez & King, 2009). For the purposes of this study 'caring' in the parental context of PPC is understood according to the conceptual definition provided by Veberne et al. (2017) as 'an expanded parenting role that involves nursing, technical and emotional tasks, such as learning about the disease, managing the child's disease, and managing one's own particular situation' (p.344).

Table 1. Categories of LLCs and LTCs, as defined by ACT/TFSL (1997, 2013)

Category	Description	Diagnostic examples
1	Life-threatening conditions for which curative treatment may be feasible but can fail.	Cancer, irreversible organ failure of the heart, liver or kidney.
2	Conditions where premature death is inevitable. Long periods of treatment may occur in an effort to prolong life.	Cystic fibrosis, Duchenne muscular dystrophy.
3	Progressive conditions without curative treatment options, where treatment may extend over many years.	Batten disease, mucopolysaccharidoses.
4	Irreversible but non-progressive conditions causing severe disability and likely premature death.	Severe cerebral palsy, brain or spinal cord injury.

Although it is understood that parents often assume a central role in caring for these children, PPC is still an emerging subspeciality (Liben, Papadatou & Wolfe, 2008). Therefore relatively few academic studies have been published within this clinical field to date (Riffin et al., 2015). At present there is a paucity of research involving LLC or LTC as more emphasis has historically been placed on the study of non life-threatening chronic illnesses (Eccleston, Palermo, Fisher & Law, 2012). A review of the literature reveals many topical articles on parenting children with diabetes (Nieuwesteeg et al., 2016) or juvenile arthritis (Yuwen, Lewis, Walker &, Ward, 2017). However, the parental experience of caring for a child with a LLC or LTC is rarely addressed (NICE, 2016) and despite recommendations for the development of a more substantive evidence base, few relevant contemporary studies exploring this phenomenon appear to exist.

Clearly, the evidence base for parental caregiving in LLCs and LTCs remains modest. Of the few existing published articles in this field, most are quantitative projects (e.g. Stuber & Shemesh, 2006), which aim to explore parents' experiences in relation to models of stress and psychopathology (Ware & Raval, 2007). Experiential qualitative studies are fewer in number (Malcolm et al., 2012). Qualitative research is considered helpful especially where 'little is known about a topic or evidence is sparse' (Somanadhan & Larkin, 2016, p.2). Additionally, this methodological approach is considered well suited to exploring individual human experiences within a health psychology framework (Smith, 2011). Qualitative research studies can be used to investigate the idiosyncratic experiences of parents and provide a more detailed understanding of their life context when supporting a child with complex illness concerns (Smith et al., 2006).

A synthesis of the qualitative literature in this area therefore appears timely and may help to advance our understanding of what it is like for parents to care for a child with a LLC or LTC. Previous qualitative review articles have tended to explore the experiences of parents caring for children with cancer (Gibbins, Steinhardt, & Beinart, 2012), particular intellectual /developmental disorders (e.g. autism; DePape & Lindsay, 2015) and non life-threatening chronic illnesses (e.g. diabetes; Alsaleh, Smith, & Taylor, 2012). Although the results from these studies enhance our ability to understand parents' experiences, the findings are often diagnosis specific, and fail to provide insight into the experiences of parents caring for children with non-cancerous LTCs (Lenton, Stallard, & Mastroyannopoulou, 2001) or non-curable LLCs (Popejoy et al., 2017). Parental caregiving has not yet been reviewed in a severe LLC and LTC context (Bally et al., 2018); although LLC/LTC prevalence is increasing (Fraser et al., 2012) and further qualitative review articles are considered a priority in this area (Ling, 2012).

To our knowledge, only one study exists that has qualitatively reviewed the experiences of families that have children with LLCs and LTCs (Bally et al., 2018). However this review did not solely focus upon parents. Wider familial experiences were examined (e.g. grandparents, siblings) and the review omitted any papers published after 2014, meaning the most recently published literature was excluded (e.g. Collins et al., 2016). The concept of caregiving was also not explicitly explored and therefore scope for a novel synthesis in the field still exists that can explore this construct exclusively in parents of children with LLCs and LTCs[1].

Aim & review question

Subsequently, the aim of this review is to draw together the relevant contemporary qualitative literature for the first time and systematically explore the parental experience of caring for a child with a LLC or LTC. Completing a novel qualitative synthesis of the research in this area may help to make the findings more accessible for practical application in clinical settings by identifying overarching themes that exist across the literature (Heath, Farre, & Shaw, 2017). The findings from this review may also be used to inform evidence-based family centered support and tailor services to meet the needs of this particular parenting population, thereby offering a guide to improve the systemic delivery of care across the PPC sector (Rempel, et al., 2013). Overall, the primary question examined is 'what are the experiences of parents when caring for a child with a LLC or LTC?'

Methods

Guidance & registration

This review followed the ENTREQ guidance for transparency in reporting qualitative research (Tong, Flemming, McInnes, Oliver, & Craig, 2012) as well as the Centre for Reviews and Dissemination (CRD) guidance for undertaking a healthcare related systematic review (CRD, 2009). The review protocol is registered on PROSPERO (Registration no: CRD42017083265,

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[1] Further differences with Bally et al. (2018) are outlined in appendix A.

registration date: 3rd December 2017), a prospective register of systematic reviews. The completed PROSPERO record is available from www.crd.york.ac.uk. This study followed an established narrative synthesis methodology described in Popay et al. (2006), which involved tabulation and thematic analysis to systematically integrate the data from included research (Popay et al, 2006).

Inclusion/exclusion criteria

Inclusion and exclusion criteria were generated *a-priori* to identify the relevant qualitative literature. Eligibility criteria for inclusion were developed using the SPIDER criteria (Sample, Phenomenon of Interest, Design, Evaluation, Research type; Cook, Smith, & Booth, 2012) for qualitative non-intervention review studies, as recommended by the Joanna Briggs Institute (2011) and the Cochrane Qualitative Review Group (2015). Exclusion criteria were generated using selected items from Gibbins et al. (2012); Smith et al. (2015).

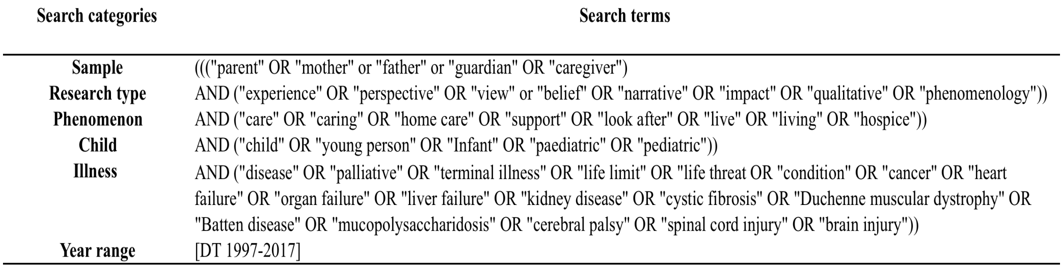
Inclusion criteria were a) qualitative b) peer review journal articles involving c) parents d) actively caring for a child aged ≤18 years e) with a LLC or LTC diagnosis f) which fell within an ACT/TFSL (1997) category (table 1). Studies were required to focus g) primarily on parents personal experiences, attitudes and perspectives towards caring for their ill child. Articles were only selected for inclusion if they presented original qualitative data (i.e. qualitative data not extracted from another pre-existing published paper). Only articles published between 1997-2017 were included to ensure a contemporary focus and consistency with the introduction of the ACT/TFSL criteria (1997,1st ed.; table 1).

Exclusion criteria were a) non-English language publications b) quantitative/mixed methods papers and c) journal articles not subjected to peer review. Studies were excluded d) if they used data obtained from non-parent family members. To reduce problems associated with impaired memory and perception e) retrospective studies involving parents of formerly ill children or bereaved caregivers were excluded. Articles were excluded if they primarily studied f) communication from HCPs regarding diagnosis/prognosis g) or non life-threatening illness. As contemporary PPC research considers equality of male and female representation a priority (Goldstein, Akré, Bélanger, & Suris, 2013) studies were excluded h) if they did not include perspectives from parents of both genders.

Information sources and search strategy

Systematic searches were conducted in Scopus, PsycINFO, the British Nursing Index (BNI), Web of Science, CINAHL, AMED and MEDLINE. Further attempts to locate published literature were made by manually searching the Health Research Authority (HRA) Register (UK), the IJS research registry (international), Google Scholar, and the children's palliative care abstract register at TFSL (togetherforshortlives.org.uk). The primary author then examined the reference lists of every eligible study for additional relevant articles as part of a further manual search for grey literature. All searches were completed on 3rd December 2017, and searched the literature from 1st January 1997 to 3rd December 2017.

The primary author (T.M.) developed a pre-planned search strategy that used both common diagnostic terminology from the ACT/TFSL categories (2013; table 1) and general text. Terms within the search were checked against evidence based search strategies published by the Cochrane Pain and Palliative Supportive Care (PaPas) review group (http://papas.cochrane .org), and were also agreed with two experienced paediatric healthcare professionals (K.M., J.Y) prior to implementation. Search terms were then peer reviewed by an information specialist at the UEA, and revised to ensure conceptual accuracy. T.M. then examined the finalised search against the PRESS 2015 electronic search strategy checklist to limit quality issues and ensure rigor (McGowan et al., 2016; Bally et al., 2018). An example image of the search strategy used for each systematic database search is provided in figure 2.



Search categories	Search terms
Sample	((("parent" OR "mother" or "father" or "guardian" OR "caregiver")
Research type	AND ("experience" OR "perspective" OR "view" or "belief" OR "narrative" OR "impact" OR "qualitative" OR "phenomenology"))
Phenomenon	AND ("care" OR "caring" OR "home care" OR "support" OR "look after" OR "live" OR "living" OR "hospice"))
Child	AND ("child" OR "young person" OR "Infant" OR "paediatric" OR "pediatric"))
Illness	AND ("disease" OR "palliative" OR "terminal illness" OR "life limit" OR "life threat OR "condition" OR "cancer" OR "heart failure" OR "organ failure" OR "liver failure" OR "kidney disease" OR "cystic fibrosis" OR "Duchenne muscular dystrophy" OR "Batten disease" OR "mucopolysaccharidosis" OR "cerebral palsy" OR "spinal cord injury" OR "brain injury"))
Year range	[DT 1997-2017]

Figure 2. Search strategy

The articles identified from each search were exported to EndNoteX8® citation manager and duplicates were removed. The first author independently screened all titles and abstracts for relevance and discarded articles that failed to meet the inclusion/exclusion criteria. Studies potentially containing relevant data were extracted and read in full by T.M. to assess eligibility. Any uncertainties regarding eligibility were then discussed within the wider research team (K.M., J.Y.) until they were resolved via consensus agreement. All remaining articles were assessed against a 4-item screening checklist developed by Caroll, Booth, & Lloyd-Jones (2012), which is designed to assist the early identification and exclusion of inadequately reported qualitative studies. Each of the remaining articles were examined against the checklist criteria and excluded from the synthesis unless they met at least three of the four pre-specified checklist criteria. Ratings were administered by the first author (T.M.) and checked by a second rater[2] for consistency.

Data synthesis

Narrative synthesis is frequently utilised to examine illness experience from an emic perspective (Vallido, Wilkes, Carter, & Jackson, 2010), and therefore this widely accepted approach was employed (Popay et al., 2006). Narrative synthesis focuses on creating a textual summary to ‘tell a story’, and aims to synthesise diverse literature (Popay et al., 2006), thus making it

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[2] A current third-year UEA trainee clinical psychologist.

suitable for outlining complex health related research material (Wiles, Cott, & Gibson, 2008). In accordance with this approach data analysis proceeded in multiple discrete stages. The relevant data were firstly extracted from each study and tabulated to create a preliminary synthesis (Popay et al., 2006). For each study the following data were tabulated: first author, publication year, location, aim, methodology, sample *N*, age range, diagnoses, data collection method and key study results. Steps were then taken to thematically analyse the results across included studies (Popay et al., 2006) using the analytical framework developed by Braun & Clarke (2006). This six-stage framework involved (1) data familiarisation (2) coding (3) theme development (4) theme review (5) agreeing the final theme labels and (6) producing a final report.

(1) Data familiarisation: The first analytical stage involved becoming familiar with the data. To ensure immersion in the data the primary author carefully examined the preliminary tabulated synthesis and re-read each included study separately. Further repeated re-readings were also undertaken as the analysis progressed to ensure the results were continually grounded in the original textual material.

(2) Coding: After becoming familiar with the data the primary author completed systematic line-by-line coding for every available text in NVivo11. To initiate the coding the results sections of each study were extracted into NVivo11. The extracted material from the results sections included all headings, participant quotations and secondary interpretations provided by the original study authors, meaning the entire results sections of the selected studies were subject to analysis. The findings were then coded inductively at an interpretative level. Codes were created by highlighting a relevant extract of text in the results section of a study and coding each extract using the code 'at new node' function in Nvivo11. Codes consisted of a brief interpretative statement that reflected the underlying meaning or essence of a particular extract. The coding process involved coding both the participants' own quotations and the authors broader interpretations within the text.

(3) Theme development: The codes were grouped together to succinctly capture similarities across included studies. The clusters of codes were relabelled at this point at a higher level of abstraction to form candidate themes (Braun & Clarke, 2006). The candidate themes were subsequently reviewed and refined through a process of further iterative relabelling, until a coherent set of themes emerged that provided a clear and comprehensive representation of the data corpus.

(4) Theme review: Each theme was assessed for homogeneity, frequency of data and relevance. Throughout this process the themes were checked repeatedly and scrutinised against the original results section from each article to ensure immersion in the data (Braun & Clarke, 2006). Themes were subjected to repeated discussions within the research team before being finalised. As part of this process the second and third authors were independently given copies of the Nvivo file and were asked to review the final themes alongside the extracts that comprised each theme. Both authors reviewed the file and verified the themes as being credible representations of the data corpus (Popay et al., 2006).

(5) Agreeing theme labels & (6) producing the final report: As a result of the theme review conducted by the second and third authors, it was agreed that the theme labels (produced at stage 3) were credible. Hence no changes were made to the names of the themes at this stage. Finally, the primary author completed a write up of the identified themes, which included use of the CASP tool to critically examine the robustness of the synthesis (Popay et al., 2006).

Quality Appraisal

Quality assessments were conducted using the 2017 qualitative CASP tool (http://www.casp-uk.net). The CASP tool contains 10 questions that allow the reviewer to assess each qualitative study in terms of its transparency and evaluative rigour (Zander, Hutton, & King, 2010). CASP ratings were calculated for every included study using a three-point scoring guideline outlined in Rushbrooke, Murray & Townsend (2014). Accordingly, 2 points were assigned if the study provided a full and detailed response to the question, 1 point for a moderately informative response or 0 points if the study provided little/no information. This scoring method is often applied within the qualitative synthesis literature (McCann, Lee & Brown, 2016; Hendry, Snowden & Brown, 2018). In line with CASP convention, questions 1-10 were scored. Hence, total possible scores range from 0-20. Studies scoring ≥17 are considered high quality (Hendry et al, 2018); studies scoring 11-16 are considered moderate quality, and those scoring ≤10 are considered low quality (Rushbrooke et al., 2014). Quality assessments were independently conducted by one rater (T.M.) and checked for consistency by the second author (K.M.). No studies were weighted or excluded based on their CASP results, as this follows recommendations from Drew, Lavy, & Gooberman-Hill (2016).

Results

Search results and study selection

Following the systematic and manual search, 767 articles were identified and exported to EndnoteX8®; 478 duplicates were removed. Titles and abstracts from 289 articles were screened and 246 were discarded as they failed to meet the inclusion/exclusion criteria. Studies potentially containing relevant data were extracted and read in full by T.M. to assess eligibility. 43 full text articles were assessed; 14 eligible articles were located. Five additional relevant articles were identified via the reference list search, meaning a total of 19 eligible articles were identified from the initial searches.

Studies have previously shown that poor reporting or thin description within included papers can adversely affect review quality (Thomas & Harden, 2008). As a result each of the 19 articles were assessed against the 4-item screening checklist developed by Caroll, Booth, & Lloyd-Jones (2012). Seven studies failed to meet the threshold for adequate reporting using this approach and were excluded; meaning a total of 12 adequately reported eligible studies were included in the synthesis. A PRISMA (2009) flow-diagram outlining the study screening and selection process is presented in figure 3.

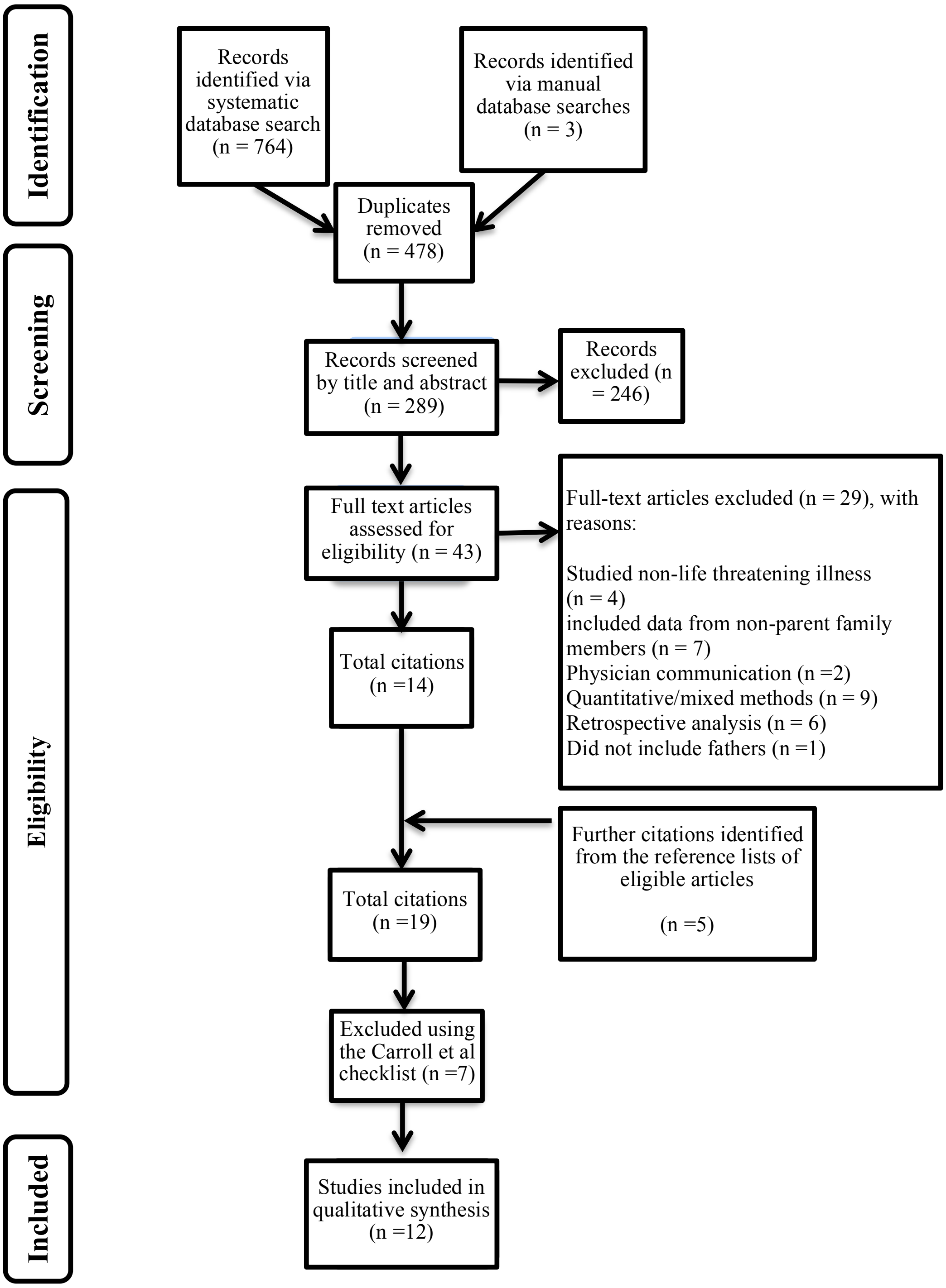


Figure 3. PRISMA (2009) flowchart

Preliminary Synthesis

12 qualitative studies were included in the review & synthesised. Study characteristics are presented in table 2. The 12 included studies described parental caregiving as being a multifaceted role (e.g. liaising with health professionals, administering medical technical procedures at home). Studies were published across 10 countries, Australia (n=3), USA (n=1), Canada (n=2), Iran (n=1), European countries (n=4), and Hong-Kong (n=1), over a 20-year period (1997-2017). Across studies participants (n=275) were typically mothers (n=195) rather than fathers (n=80), caring for children (n=193) with heterogeneous illness conditions falling into one of the four ACT/TFSL disease categories (table 1), including cancer (n=116), cerebral palsy (n=40), congenital anomalies (n=11) mucopolysaccharidosis (MPS; n =8), degenerative neurological conditions (n=4), metabolic disease (n=2), central congenital hypo ventilation syndrome (n=1), multicystic bilateral renal dysplasia (n=1), cystic fibrosis (n=1), Schwartz-Jampel syndrome (n=1), Sandhoff disease (n=1), nemaline rod myopathy (n=1), Lennox Gastaut syndrome (n=1), Duchenne muscular dystrophy (n=1), spinal muscular atrophy type II (n=1), metachromatic leukodystrophy (n=1), Retts syndrome (n=1) and respiratory distress (n=1). Semi structured interview designs were chosen to study caregiver experiences in all studies (n=12), although focus groups were used to supplement the interview content in one instance. Analytical strategies included thematic analysis (n= 4), grounded theory (n=5) and phenomenological approaches (n=3).

Table 2. Characteristics and results of included studies (n=12)

First author, publication year	Location	Parent *N*	Aim	Method	Age range for child	Age range for parent	Diagnoses	Data collection method	Key study results
Collins (2016)	Australia	14	"To provide an in-depth exploration of the prevalent lived experiences of parents who are currently providing care for a child with a life-limiting condition in Australia" (P.951).	Thematic analysis	3 months - 17 years	25–51 years	Cancer (metatastic pleomorphic, xanthoastrocytoma, leukaemia) central congenital hypo ventilation syndrome, multicystic bilateral renal dysplasia, cystic fibrosis, Schwartz-Jampel syndrome,, Sandhoff disease, degenerative neurological condition, serious undiagnosed neurological disability, nemaline rod myopathy, Lennox Gastaut syndrome, severe cerebral palsy and associated complications.	Semi structured interview	Parents reported physical/social isolation from the community, assuming ownership of the caring duties, and living with the likelihood of the child dying. Parents also derived meaning and purpose from their caregiver role.
Davis (2010)	Australia	37	"To explore the quality of life of mothers and fathers caring for children with cerebral palsy aged from 3 to 18 years" (P.64).	Grounded theory	3-18 years	not stated	Cerebral palsy.	Semi structured interview	Caring for a child with cerebral palsy can negatively affect parents' sleep, marital relationships and social relationships. New social relationships were developed via interactions with other parent caregivers.
Flury (2011)	Switzerland	12	"To explore the experiences of parents who care for their child, newly diagnosed with cancer, after the initiation of treatment and the first hospital discharge from 2 Swiss children's hospitals" (P.144).	Thematic analysis	2 - 16 years	23-49 years	Cancer conditions (e.g. acute lymphoblastic leukemia, rhabdomyosarcoma, and non-Hodgkin's lymphoma).	Semi structured interview	Delivering care at home required parents to learn numerous new tasks and parents often felt they were in an perpetual state of crisis. Parents often struggle to retain employment due to caregiving demands.
Hayles (2015)	Australia	13	"To explore parents' experiences of healthcare/ delivering care for their children with cerebral palsy living in a regional area of Australia" (P.1).	Grounded theory	22 months -16 years	not stated	Cerebral palsy.	Semi structured interview & focus groups	Parents described caregiving as a stressful time in which the childs needs were constantly changing. Parents had difficulty navigating the healthcare system due to multiple organizational and access issues.
Somanadhan (2016)	Republic of Ireland	8	"Aimed to understand and interpret parents' experience of living and caring for a child with MPS" (P.2).	Thematic analysis	6 months - 22 years	not stated	MPS I syndromes (Hurler syndrome, Scheie syndrome), MPS II (Hunter syndrome), MPS III (Sanfilipo syndrome) and MPS VI (Maroteaux-Lamy syndrome).	Semi structured interview	Parents described life as emotionally challenging and characterised by uncertainty. Parents described healthcare provision as fragmented and unsatisfactory. Parents benefited from networking with other families.
Verberne (2017)	Netherlands	42	"Aimed to provide a comprehensive overview of parental caregiving, based on the lived experience of parents caring for a child with a life limiting disease" (P.344).	Thematic analysis	0-over 16 years	under 30 to over 40 years	Congenital anomalies, neurodegenerative disease, metabolic disease, cancer (central nervous system tumour, bone/soft tissue sarcoma, neuroblastoma, leukaemia).	Semi structured interview	Parents constantly worked to manage and relieve disease symptoms. Parents felt it was difficult to balance multiple demands on their time. Care systems were considered difficult to navigate.

First author, publication year	Location	Parent *N*	Aim	Method	Age range for child	Age range for parent	Diagnoses	Data collection method	Key study results
Taleghani (2012)	Iran	15	"Aimed to gain an understanding of the lived experiences of parents of children diagnosed with cancer, and to examine their descriptions of the situations they found themselves in" (P.342).	IPA	2-12 years	25-45 years	Cancer.	Semi structured interview	Parents experienced a psychological burden when caring for their child. Seeing other parents in similar circumstances helped however, as did spirituality and religious belief. Parents felt isolated from society due to caregiving demands.
Kars (2008)	Netherlands	23	"Aimed to explore the lived experience of parenting the child with leukaemia during the first year after diagnosis" (P.1555).	Grounded theory	2-12 years	29-42 years	Cancer (lymphoblastic leukaemia).	Semi structured interview	Protection and preservation were core aspects of 'being there' for the child. Emotional support for the parent is helpful. Parents often advocated for the childs interests. Parents experienced emotional difficulties due to the demanding nature of their role.
Klassen (2012)	Canada	76	"Aimed to ask parents of children with cancer about areas of their health (physical, psychological, and social) that had been compromised due to caregiving as well as any positive impacts of caregiving" (P.1596).	Grounded theory	3 months - 17 years	22-61 years	Cancer (leukemia, lymphoma, sarcoma, neuroblastoma, Wilm's Tumor, brain cancer, liver cancer).	Semi structured interview	Parents described sleep difficulties, anxiety, low mood, and social isolation. Positive impacts were also reported including patience, inner strength, and new life perspectives. Parents also endeavoured to share information with other parents facing similar difficulties.
Patterson-Kelly (2011)	USA	15	"Aimed to understand how parents who no longer live together make treatment decisions for their child with cancer" (P.2).	Grounded theory	15 months - 17 years	20-50 years	Cancer (acute lymphoblastic leukemia, brain tumor, Hodgkin's lymphoma, and neuroblastoma)	Semi structured interview	Disease management exacted an emotional toll on caregivers. Parents felt it was difficult for anyone else to understand their situation.Treatment decision making was frequently undertaken by the custodial parent.
Wong (2006)	Hong Kong	9	"Aimed to understand the coping experiences of Chinese parents with children having cancer during the treatment stage" (P.711).	Colaizzi's (1978) phenomenological methodology	9 months-15 years	31-45 years	Cancer (osteosarcoma, Ewings Sarcoma, leukemia, neuroblastoma, retinoblastoma, Rabdomyo-sarcoma).	Semi structured interview	Parents go through various stages of coping. Emotional support was considered important for parents.They wished to discuss their experiences with other parents caring for children with cancer. Peer support was considered highly beneficial, particularly during the early stage of treatment.
Gravelle (1997)	Canada	11	Aimed to understand how parents manage their childs illness "during the complex chronic phase" (P.738).	Omery's phenomenological method	26 months-16 years	not stated	Duchenne muscular dystrophy, spinal muscular atrophy type II, metachromatic leukodystrophy, Retts syndrome, cerebral palsy with microcephaly, and respiratory distress.	Semi structured interview	Facing adversity was defined as the central theme. Parents encountered multiple challenges due to caregiving. Parents felt frustrated and angry with the beuracracy involved in the heathcare system. Families needs change constantly due to the evolving nature of the childs condition.

Detailed Synthesis

Through the synthesis conducted within Nvivo five emerging themes were identified; ‘navigating the system’; ‘burden of care’; ‘living with uncertainty’; ‘strength through adversity’ and ‘connecting with other families’. Each theme uniquely contributed to explaining the parental experiences of caring for a child with a LLC or LTC. Frequency counts, identifying the number of studies including each theme, are reported in table 3.

Table 3. Frequency counts indicating the presence of themes across included studies[i]

Study (first author)	navigating the system (11/12)	burden of care (12/12)	living with uncertainty (9/12)	strength through adversity (11/12)	connecting with other families (10/12)
Collins (2016)	•	•	•	•	•
Davis (2010)	•	•	•	•	•
Flury (2011)	•	•	•	•	•
Hayles (2015)	•	•	•	•	•
Somanadahan (2016)	•	•	•	•	•
Veberne (2017)	•	•	•	•	
Taleghani (2012)		•		•	•
Kars (2008)	•	•		•	
Klassen (2012)	•	•	•	•	•
Patterson-Kelly (2011)	•	•			•
Wong (2006)	•	•	•	•	•
Gravelle (1997)	•	•	•	•	•

[i] table style is derived from Heath et al. (2017).

Navigating the system

The theme ‘navigating the system’ is represented across 11 of the 12 articles. The studies that spoke most directly to the essence of this theme included an appropriate design and clearly outlined the data collection approach (table 4; Davis et al., 2011, Gravelle, 1997; Veberne et al., 2017; Somanadhan & Larkin, 2016), although one study did not account for ethical issues or describe the analytical procedure in detail (Gravelle, 1997). The theme itself described the various difficulties encountered by parents when trying to identify and obtain professional help from healthcare providers. A multitude of organisational systems with different eligibility requirements made it harder for parents to easily locate appropriate services for their child (Somanadhan &

Larkin, 2016; Davis et al., 2010). The limited accessibility and availability of different services and information meant parents had to advocate tirelessly on their child's behalf to ensure they received the right kind of medical support (Flury, Caflisch, Ullmann-Bremi, & Spichiger, 2011; Hayles Harvey, Plummer, & Jones, 2015; Patterson-Kelly & Ganong, 2011). Efforts were often expended filling in paperwork or negotiating with funders without any firm guarantee that the child's immediate healthcare needs would be met (Gravelle, 1997). Accordingly, parents described the process of navigating the system as a 'fight' or 'battle' for resources:

'It's been a constant fight and battle. I'm still fighting to get a standing frame in the school…then it was a constant battle of fighting for therapy…it doesn't matter what we do, we have to fight and challenge everyone on everything' (Hayles et al., 2015, p. 8).

This metaphor extended to an understanding that parental quality of life would be enhanced if the fight could be more easily overcome:

'I think for us probably having better access or even an access to OT would be great. To not have to feel like we were fighting for all our services all the time, and to have that…what would have made our lives easier in retrospect' (Davis et al., 2010, p.70).

Negotiating access to a service was seen as important but stressful (Davis et al., 2010). As the child aged, parents were often forced to seek out new services for the child. Hence, navigating the system was conceptualised as a perpetual struggle for resources that continually evolved and required constant parental attention over time (Klassen et al., 2012). Moving to a new school or town often meant a change in service provider, resulting in further bureaucracy, and the need to navigate a plethora of new systems with divergent referral criteria (Hayles et al., 2015). In this regard parents came to think of the healthcare system as inconsistent and impersonal (Somanadhan & Larkin, 2016). One parent likened it to a revolving door:

'So you go through, it is like a revolving door, you go in, you go out, you go in, you go out, and you are only the number of the day, and the doctor and the patient can make it special. But to the system, I don't think it is a caring enough system' (Somanadhan & Larkin, 2016, p.9).

Moving from infancy through to adolescence often meant searching for a different service provider, managing further bureaucracy, and continuing to fight for adequate provision. Over time, this impacted upon parental confidence in the system. Frequently, parents stated that they were in a better position than the medical profession to deliver care attuned to their child's needs (Collins et al., 2016; Veberne et al., 2017; Patterson-Kelly & Ganong, 2011). They felt that unlike themselves healthcare providers did not always place the child first (Kars, Duijnstee, Pool, Van Delden, & Grypdonck, 2008). For some parents this shortcoming meant that they expressed a desire to 'walk away' and stop navigating the system entirely (Somanadhan & Larkin, 2016). Others expressed a strong commitment to assume almost total responsibility for managing the child's care needs:

'I regarded myself as the key caregiver and I could not escape from my responsibility. I tried to simplify my personal life and concentrate all my energy to meet the care demands' (Wong & Chan, 2006, p.714).

Burden of care

The theme 'burden of care' is represented in all 12 articles. Six of the papers that supported the theme were deemed to be of high quality based on their clear in-depth description of the analysis procedure and detailed statement of the research findings (table 4; Collins et al., 2016; Flury et al., 2011; Hayles et al., 2015; Verberne et al., 2017; Wong & Chan, 2006; Paterson-Kelly & Gangong, 2011). The theme itself refers to the multitude of care tasks undertaken by the parent. Parents came to consider themselves experts in the management of disease by delivering direct 'hands on' care to the child. The diverse medical tasks completed by parents included antibiotic prophylaxis (Flury et al., 2011), administering chemotherapy (Taleghani, Fatizadeh, & Naseri, 2012), purchasing hoists (Somanadhan & Larkin, 2016), controlling seizures (Collins et al., 2016) monitoring side effects (Veberne et al., 2017), giving tube feeding (Klassen et al., 2012) and modifying the child's diet (Wong & Chan, 2006). These procedures were adjusted depending on the child's age and physical health status to maximise comfort and ensure the child complied with various treatment regimes throughout the day (Verberne et al., 2017; Davis et al., 2010; Hayles et al., 2015).

Due to the demands associated with providing various care tasks, time became a limited resource (Flury et al., 2011). Parents were keen to stress that the provision of care was extremely time consuming (Kars et al., 2008). It represented a 24-hour per day responsibility from which the parents could not easily extricate themselves without feeling guilt (Veberne et al., 2017). Parents described the substantial amount of time they expended caring for their child:

'And it's very time-consuming. You know, just showering (our child) takes 45 min. And you have to plan all that to try and fit everything in.'(Davis et al., 2010, p.68)…'I feel quite alone and overwhelmed with this caring role that seems like a life sentence, or solitary confinement' (Collins et al., 2016, p.954).

Another parent of a child with cerebral palsy (aged 12) compared the child's care needs to a newborn infant. She reflected on the burden this imposed:

'*You could say…it's like you've got a baby 24 h a day, like when a baby is newborn or before they reach 6 months or whatever and they can't really do things for themselves…it's just full on 24 h a day'* (Davis et al., 2010, p.68).

Restrictions due to a lack of time meant many parents described their seclusion from the wider community (Collins et al., 2016; Taleghani et al., 2012). They became isolated from former friends or exhibited a growing reluctance to socialise as fatigue set in (Gravelle, 1997). In many cases parents also found it harder to maintain employment (Flury et al., 2011, Davis et al., 2010). Regular work hours were considered incompatible with the requirement to manage the child's immediate needs and respond to particular care demands at short notice (Flury et al., 2011; Klassen et al., 2012):

'I've had to give up work to care for her. I hate it because it was the only thing for me. You know, I was me, I wasn't mum…losing that was huge…I can only commit to a few hours a week…so I'm just doing some voluntary work to get me out of the house. I don't want to be defined by being a carer' (Collins et al., 2016, p.954).

Over time, the burden of providing perpetual and prolonged care led to emotional health problems. Parents reported severe stress (Davis et al., 2010), depression (Wong & Chan, 2006), anxiety (Paterson-Kelly & Gangong, 2011; Somanadhan & Larkin, 2016) and anger (Klassen et al., 2012). For parents of children with LLCs these emotional responses were further complicated by anticipatory grief relating to the child's impending death (Collins et al., 2016; Gravelle, 1997).

Living with uncertainty

The theme 'living with uncertainty' is represented by 9 of the included articles. The articles that strongly represented this theme provided less consideration of reflexivity, but stated clear aims, a design appropriate for the research question and used an appropriate qualitative methodology (Klassen et al., 2012; Somanadhan & Larkin, 2012). The theme itself refers to the uncertainty felt by parents about the child's condition and its impact on the child's future (Collins et al., 2016; Flury et al., 2011). Parents of children with both LLCs and LTCs were aware that the child's condition could worsen at any time (Somanadhan & Larkin, 2016). A fear of the future persisted because even during periods of relative stability there remained a strong possibility that the child's physical health status would change rapidly and deteriorate further (Veberne et al., 2017; Klassen et al., 2012; Wong & Chan, 2006). Parents of children with progressive conditions described a sense of continual unease because the child's care needs were constantly evolving over time:

'It changes over the years. The child we have now…is a completely different child that we had when our child was 3…Yes, the condition is probably less mentally draining now and more physically challenging'. '...So we probably feel a little more vulnerable now, and a little more shook up because the condition has very gradually changed from day one' (Somanadhan & Larkin, 2016, p.10).

For this reason parents described being in a position of ambiguity where they were unsure what might happen next (Davis et al., 2010). In order to try and manage this uncertainty parents lived moment by moment (Hayles et al., 2015). They shifted from long-term care planning to short term planning as they tried to limit the impact of uncertainty (Gravelle, 1997). Parents also sought information about life expectancy and survival rates in order to estimate the amount of time they had left with their child, although in one case this seemed to increase uncertainty further:

'In Australia, 600–700 people will die waiting for a kidney transplant. But, does that include the people who aren't active on the list? This stuff is always in the back of your mind. I don't know how much time I've got left with my little girl' (Collins et al., 2016, p.955).

Due to the uncertain nature of the child's condition parents felt that they were constantly learning in response to new challenges that presented themselves during the illness life course. Parents learnt to adapt their lives in order to manage the child's difficulties, however they also admitted to a process of 'learning on the go', in which they reacted to new needs and complexities as they emerged.

'Each time something happened…we learnt by it....No one told us what to expect…we just winged it each year, by the year' (Hayles et al., p.5).

This also led them to predict further challenges that might arise in the context of the child's developmental trajectory during puberty:

'See, we're about to go through another change where (child)'s hormones are going to come on-board and she will.go through puberty. . .that's another big deal for us to have to go through, and need direction and help with from our support services' (Hayles et al., p.5).

Strength through adversity

The theme 'strength through adversity' is represented in 11 of the included articles. Whilst high quality articles were included within this theme (table 4; e.g. Verberne et al., 2017), a number of the articles that prominently articulated this theme lacked sufficient quotations from participants to support their analysis (Gravelle et al., 1997; Taleghani et al., 2012) or did not account for the potential impact of researcher bias in the analysis (Klassen et al., 2012). The theme itself refers to the way that parents were able to maintain a positive outlook despite their child's LLC or LTC (Veberne et al., 2017; Gravelle, 1997; Hayles et al., 2015; Kars et al., 2008). Parents often felt their position contrasted favourably with other families caring for children with the most severe types of progressive conditions (Somanadhan & Larkin, 2016; Davis et al., 2010; Wong & Chan, 2006). By reflecting on this difference, parents of children with LTCs realised that their child's life prospects could be considerably worse. One parent of a child with MPS explained this position:

'And you see other parents, I mean the MPS disorder is horrendous, certain ones, Hurler's, Sanfilippo, Hunter's, I mean we have lost so many children. And they have a short life expectancy. We feel luck that it is not cancer, that is it not Sanfilippo, that it is not Hunter's, so that is probably a way of coping. We have probably taught ourselves to cope that way' (Somanadhan & Larkin, 2016, p.7).

Parents also obtained strength through adversity by redefining expectations for themselves and their child (Flury et al., 2011). After diagnosis many parents came to realise the importance of celebrating small goals or achievements. Long term expectations they previously had for the child, such as getting married or achieving financial success, were replaced by a more immediate emphasis on having fun and maintaining quality of life:

'I hope my child will fully recover and resume school soon. It is not important whether he has good academic performance, can make money or get married. All I want is for him to be healthy' (Wong & Chan, 2006, p.715).

Parents felt that caring for their child had given them existential insight, life satisfaction, and a new ability to prioritise what is truly meaningful in life (Collins et al., 2016). Through managing adversity parents developed a deeper relationship with the child. They described the development of a closer bond, characterised by love, warmth, empathy, respect and a mutual appreciation of time spent together (Kars et al., 2008). Parents explained that having supported their child in adverse circumstances they were more resilient (Collins et al., 2016), more adaptable (Veberne et al., 2017), more empathic (Klassen et al., 2012) more spiritual (Taleghani et al., 2012), and better able to manage any potential challenges they might face in the future. As one mother summarised:

'When something like this happens to your family, it can devastate you, or it can have a positive effect...It's made me a much stronger person, a much more caring person... [I have] a better understanding of what other people are going through...a better understanding of life, I guess' (Gravelle, 1997, p.741).

Connecting with other families

The theme 'connecting with other families' is represented in 10 of the included articles. The studies that prominently supported this theme did not always consider reflexivity in the formulation of the research question or methods (Somanadhan & Larkin, 2016; Taleghani et al., 2012), although a number provided a clear in-depth statement of their findings (Wong & Chan, 2006; Davis et al., 2010; Collins et al., 2016). The theme itself refers to the way that parents felt that they benefited from the opportunity to share their experiences with other mothers and fathers caring for children with similar illnesses (Collins et al., 2016; Davis et al., 2010; Flury et al., 2011; Hayles et al., 2015;

Klassen et al., 2012; Taleghani et al., 2012). Parents emphasised the importance of interacting with these families on a regular basis (Taleghani et al., 2012; Wong & Chan, 2006). Talking at support groups or on the hospital ward provided a way for parents to bond with other carers, and constituted a powerful way to receive emotional support:

'Whenever I sit there to cry, my roommates console me by saying there are worse diseases than this and a lot of people lose their children. They console me' (Taleghani et al., 2012, p.344).

Others felt that they benefited from the opportunity to obtain additional information about aspects of care:

'There'd be a group of parents sitting on the side…and we'd start talking…'oh, did you hear about this?' or, 'did you know that you could do that? We teach each other because nobody's teaching us' (Hayles et al., 2015, p.6).

While mothers often benefited from this type of information sharing, certain marginalised groups did not always receive the same opportunities. Immigrant parents often relied on family support (Klassen et al., 2012), while some fathers felt that they were minimally involved with other parents (Patterson-Kelly & Ganong, 2011):

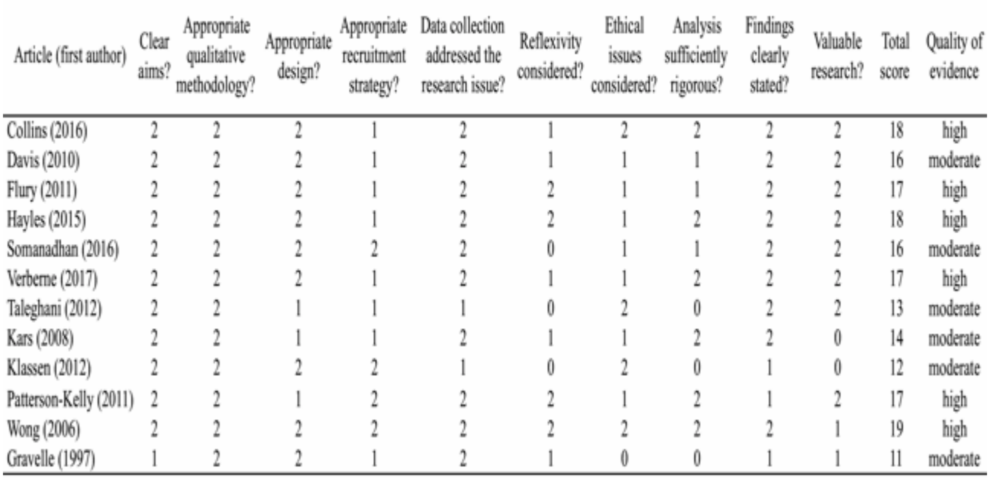
'And from a father's point of view there's nothing, you know (my wife) goes to early intervention programs all the time…90% of fathers have to go out and earn a living. So I don't get together with other fathers with other disabilities and so forth and vent…for a father there's absolutely nothing' (Davis et al., 2010, p.69).

Parents of children with rare conditions tended to also underline the importance of interacting with others online, as it was not always possible to find similar parents living nearby (Hayles et al., 2015). The Internet provided a way to stay in touch with the outside world in the context of the care demands imposed by the child's illness (Klassen et al, 2012; Somanadhan & Larkin, 2016).

Quality Appraisal

CASP ratings are presented in table 4. The CASP results indicate that the included studies typically described their aims, methods, recruitment strategy and findings with clarity. All 12 studies provided a clear statement of aims and used an appropriate qualitative methodology. The research design, recruitment strategy and data collection method were outlined in 12 studies, although only 9 studies explicitly considered reflexivity. In three studies, researchers did not consider their own role and potential to exert bias through recruitment, data collection or the formulation of the research question (Klassen et al., 2012; Somanadhan & Larkin, 2016; Taleghani et al., 2012). Practices designed to maintain ethical standards were clearly documented in 11 studies. Only one study did not mention ethical issues (Gravelle, 1997). Data analyses were only considered sufficiently rigorous in 9 studies. The remaining studies did not provide an in depth description of the analysis procedure (Gravelle et al., 1997), lacked sufficient participant quotations to support their analysis (Gravelle et al., 1997; Taleghani et al., 2012) or did not account for the researchers role, potential biases and influence during analysis itself (Klassen et al., 2012). All 12 studies provided a clear statement of their research findings and 10 studies were deemed to have contributed valuable findings with clear implications for practice.

Table 4. CASP ratings across the 12 included studies



Article (first author)	Clear aims?	Appropriate qualitative methodology?	Appropriate design?	Appropriate recruitment strategy?	Data collection addressed the research issue?	Reflexivity considered?	Ethical issues considered?	Analysis sufficiently rigorous?	Findings clearly stated?	Valuable research?	Total score	Quality of evidence
Collins (2016)	2	2	2	1	2	1	2	2	2	2	18	high
Davis (2010)	2	2	2	1	2	1	1	1	2	2	16	moderate
Flury (2011)	2	2	2	1	2	2	1	1	2	2	17	high
Hayles (2015)	2	2	2	1	2	2	1	2	2	2	18	high
Somanadhan (2016)	2	2	2	2	2	0	1	1	2	2	16	moderate
Verberne (2017)	2	2	2	1	2	1	1	2	2	2	17	high
Taleghani (2012)	2	2	1	1	1	0	2	0	2	2	13	moderate
Kars (2008)	2	2	1	1	2	1	1	2	2	0	14	moderate
Klassen (2012)	2	2	2	2	1	0	2	0	1	0	12	moderate
Patterson-Kelly (2011)	2	2	1	2	2	2	1	2	1	2	17	high
Wong (2006)	2	2	2	2	2	2	2	2	2	1	19	high
Gravelle (1997)	1	2	2	1	2	1	0	0	1	1	11	moderate

Discussion

This systematic review explored the qualitative literature from 1997-2017 concerning parents' experiences of caring for a child with a LLC or LTC. A narrative synthesis of 12 studies from 10 countries, involving 275 parents, led to identification of five overarching themes; (a)'navigating the system'; (b)'burden of care'; (c)'living with uncertainty'; (d)'strength through adversity' and (e)'connecting with other families'. Prior existing published review articles have explored the experiences of parents caring for children with non life-threatening illnesses (Coffrey, 2006), particular intellectual/ developmental disorders (DePape & Lindsay, 2015) or cancer (Klassen et al., 2007). To our knowledge, this is the first systematic review to directly explore parents' experiences of caring for a child with a LLC or LTC using only qualitative research and narrative synthesis methods. Previous reviews have tended to adopt a disease specific approach to the study of illness (e.g. Tong, Lowe, Sainsbury, Craig, 2008; Grootenhuis & Last, 1997) while this review explored a much broader range of conditions, covering each of the four ACT/TFSL disease categories (table 1).

Findings indicated that parents generally have difficulty navigating the healthcare system effectively. Considerable ongoing effort is expended searching for appropriate services, although parents often experience problems accessing high quality service provision. Parents caring for children with a LLC or LTC encounter high levels of burden, stress and feelings of uncertainty. Despite these difficulties they are often able to maintain a positive outlook and encounter additional positive emotional changes (e.g. increased empathy). Many parents also feel that they benefit from speaking to other mothers and fathers caring for children with similar illnesses.

By synthesising the results from studies involving different LLCs and LTCs, a number of common experiences were identified. In keeping with the wider paediatric psychology literature (Eccleston et al., 2012), parents were engaged in a 'fight' for resources and encountered troubling emotions in the context of their caregiver role (Yuwen et al., 2017). Negative experiences and burden were reported in all studies, a finding which broadly supports previous research involving parents of children with chronic illnesses (Barlow & Ellard, 2006) and long-term conditions (Smith et al., 2015). Poor psychological adjustment and stress are also commonly reported in studies of parents caring for children with cancer (Grootenhuis & Last, 1997), although this review appears to indicate that these problems are not diagnosis specific. Findings were widely applicable to parents of children with various illnesses. This paper therefore illustrates a commonality of experience and a sense of shared adversity that parents may face across child illness.

In contrast to previous studies focusing on parental distress and psychopathology (Knapp, Madden, Curtis, Sloyer, & Shenkman, 2010) this paper also identified a number of positive themes. Parents exhibited growth alongside distress, as observed within the themes ‘strength through adversity’ and ‘connecting with other families’. Few articles have previously documented the positive experiences that occur for parents in the context of a LLC or LTC. Yet the findings from this study align with a strengths related account of post-traumatic growth (Tedeschi, 2011), by demonstrating that parents also exhibit positive emotional change and transformation in the context of their child’s condition.

Uncertainty and post-traumatic growth were also reported in Bally et al (2018). While the findings add credibility to those reported in Bally et al. (2018) the current study updates the results by including research published after 2014 and excluding low quality findings (Caroll et al., 2012). Additionally this study identified a number of novel themes (navigating the system, burden of care, connecting with other families) and contemporaneous articles that were not reported in Bally et al., 2018, thus adding richness and value to our understanding of parents experiences in this area (Collins et al., 2016). In focusing exclusively on parents’ experiences and the construct of caregiving this review arguably also provides a more fine-grained analysis of results in the field compared to Bally et al., 2018. However, results should also be interpreted with caution. A minority of studies did not provide in-depth descriptions of reflexivity (e.g. Klassen et al., 2012; Somanadhan & Larkin, 2016; Taleghani et al., 2012), or lacked rigour in their description of ethics, data analysis and study implications (Gravelle et al., 1997; Taleghani et al., 2012; Klassen et al., 2012).

Strengths & limitations

The review employed a robust methodology. Findings were provided from 12 contemporary peer-reviewed publications, and this study utilised a replicable procedure (figure 3), applying criteria from ENTREQ (Tong et al., 2012) and the CRD (2009) to enhance reliability. Inadequately reported studies were excluded at the point of study selection and screening. Hence, many of the 12 included studies produced high ratings against the CASP quality criteria (table 4). Expert discussions with PPC clinicians (K.M, J.Y.) and independent appraisals of the synthesis were also completed at various stages of the analysis to improve the rigour of this review study (Disler et al., 2014).

Parents' experiences were investigated across conditions. A notable strength of this review is that the studies included a number of different diagnoses and types of care requirements (Verberne et al., 2017). This review also included rare under-researched conditions by studying parents of children with MPS (Somanadhan & Larkin, 2016) and other less common LLCs (Davis et al., 2010). Accounts from these parents are usually excluded in review studies where a disease specific approach is adopted (Malcolm et al., 2012).

Search terms were collaboratively developed and refined with involvement from the wider study team. Although systematic and manual searches were conducted across 11 databases, the author cannot be certain that all relevant studies were captured. LLCs and LTCs constitute over 300 diagnoses (Hain, Devins, Hastings, & Noyes, 2013); therefore it is possible that additional eligible articles were not identified and included. Additionally, exclusion criteria were strict, leading to the omission of mixed methods papers (Worthen, Leonard, Blair, & Gupta, 2015) and those papers including only mothers or fathers (Ware & Raval, 2007). The inclusion of these papers could have yielded additional data and contributed to a richer synthesis.

Parents' experiences were investigated across conditions. However, parents were predominantly caring for children with cancer and other category 1 conditions. The results are less applicable to other rare conditions in categories 2-4 (table 1). Furthermore, a number of articles focused on mothers' accounts in a western, English-speaking context (Flury et al., 2011; Hayles, Harvey, Plummer, & Jones, 2015). Hence, a limitation of this article is that the results may not generalise as well to fathers, or parents caring for children in a non-western context. There is also a need to exercise caution in the interpretation of the findings. The CASP results indicated that certain studies lacked rigour in their description of reflexivity, ethics, data analysis and study implications.

Clinical implications

The review findings have a number of clinical implications. Firstly, detailed information is needed to help parents of children with LLC or LTC navigate systems of support. Optimal support should encompass the whole trajectory of illness, with clear information about available services being communicated regularly from the point of diagnosis onwards. Ongoing consultation and education is also needed to ensure parents are made aware of appropriate healthcare services for their child. Secondly, given the considerable burden parent's face arrangements should be put in place to assist parents with the practical aspects of delivering personal care. Practical support and guidance should be extended to help parents deliver direct 'hands on' care to the child and reduce community isolation.

Thirdly, holistic care involving psychology support may be particularly beneficial for anxious parents who are living with the uncertainty of their child's condition or experiencing other emotional stressors resulting from the demands of care provision.. Short-term goal setting and working collaboratively with the parent to redefine their expectations for the child may also further reinforce growth, resilience and recognise their capacity for meaning focused coping.

Lastly, parents may benefit from additional opportunities to meet with other mothers and fathers caring for children with similar illness conditions. Mutual support involving caregivers has previously shown to help families improve their problem solving skills, maintain their emotional health, and deliver more effective care (Wong & Chan, 2006). The findings within this study also appear to support the assertion that peer support is of practical and emotional value for parents.

Research implications

Cancer and other malignant category 1 conditions were highly represented across included studies (table 2). Future research should explore parents' experiences of caring for a child with a non-cancerous LLC or LTC. Although rare conditions were included in the review (e.g. MPS), evidence in the literature is limited for these illnesses, and additional studies are needed to assess the symptom management challenges they present (Malcolm et al., 2012). The impact of LLCs and LTCs on fathers from a non-western, non-English speaking background is largely unclear at present (Ware & Raval, 2007), suggesting there is a need to further explore the experiences of this population within a qualitative framework. In the present review the CASP results indicated areas for research improvement. Particular studies lacked sufficient participant quotations, did not account for potential researcher biases during analysis, and did not always discuss ethical issues and practice implications. Further research is therefore required to address each of the methodological limitations described.

Conclusion

This study explored parents' experiences of caring for a child with a LTC or LLC. Although parents' perspectives were explored across a number of different conditions, five consistent themes were identified within the literature. Themes reflected the fact that parents encounter both negative and positive experiences while caring for a child with a LTC or LLC. There is evidence that parents could benefit from greater nursing and psychological support to manage the demands of care provision, although additional future interventions should also aim to recognise parents existing capacity for positive emotional change and growth.

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Conflict of interest/disclosure statement

No conflict of interest is reported.

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2

Bridging Chapter

The bridging chapter (chapter 2) considers the systematic review findings (Chapter 1) and associated theory. Pennebaker's (2000) emotional disclosure theory[3] is discussed in light of the review. The theory presented is then used to provide a rationale for the Empirical Research Paper (ERP; Chapter 3) – a qualitative study that explores storytelling among parents of children with a LLC and LTC.

Parents' Experiences

Caring for a child with LLC or LTC can be a complex, emotionally demanding life experience, which often has a myriad of consequences for parental health and wellbeing (Ling, 2012). Parents are typically the main care providers, delivering 24-hour support within the family home (Remedios et al., 2015). Their burden is considerable, and the resulting stress has been recognised as clinically problematic (Rodriguez & King, 2009). In the literature it is well understood that parents of these children are particularly likely to encounter anxiety (Grant et al., 2013), fatigue (Emond & Eaton, 2004), depression (Knapp et al., 2010) insomnia (Jones, 2012) and reduced health-related quality of life (Klassen et al., 2012), while the associated physical impacts may also lead to declining health (Steele, 2016).

Research in recent years has focused primarily on the experiences of parents and the difficulties they encounter with an ill child, especially regarding the day-to-day management and treatment of child illness (D'Urso, Mastroyannopoulou, & Kirby, 2017). A significant number of studies have sought to describe the emotional turmoil experienced by parents when

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[3] Terminology derived from Schenker et al. (2015).

supporting a child with complex palliative needs (e.g. Veberne et al., 2017). Meanwhile, the neutral or positive dimensions of this life experience are rarely reported upon (Coombes, Woodward, & Norton, 2017). As the stress and burden associated with caring for a child with LLC or LTC is often emphasised (Davies et al., 2004), the majority of research in this field has focused heavily on posttraumatic stress symptoms (PTSS) and post-traumatic stress disorder (PTSD) in mothers and fathers (Vernon et al., 2017; Hoven et al., 2017), while the experience of positive psychological change and growth is rarely examined (Ljungman et al., 2014).

Although studies of benefit finding are rarely evidenced in the literature, many parents of children with LLC and LTC also report positive changes during the illness life course (Siden & Steele, 2015). In-depth qualitative studies have shown that positive feelings about caregiving often emerge in the context of these illnesses (Ware & Raval, 2007). Parents report greater adaptability (Cadell, Kennedy, & Hemsworth, 2012), maternal strength (Melnyck et al., 2004), life satisfaction (Flury et al., 2011), and exhibit improved familial communication/relationships (Patterson-Kelly & Ganong, 2012) in response to the demands of providing LLC/LTC care (Picoraro, Womer, Kazak, & Feudtner, 2014). These outcomes are evident in the systematic review (Chapter 1), and alongside other qualitative studies (Sleigh, 2005) demonstrate adaptive potential in parents of children with LLC and LTC.

Theoretical framework

Whilst parents of children with LLC and LTC often encounter positive emotional change and growth in their caregiver role (chapter 1), the reasons for this remain largely unclear (Picoraro, et al., 2014). At present there are relatively few published studies exploring the factors that directly contribute to positive outcomes in this trauma-exposed population (Cianfaglione et al., 2015). Detailed explanations of the distal and proximal factors associated with parental benefit finding are rarely provided in the PPC corpus at present, particularly as parental caregiving is rarely studied from a non-pathologising perspective (Ljugman et al., 2014), and is an emerging subspeciality in the wider paediatric psychology field (Thomas, Phillips, & Hamilton, 2018).

Authors have suggested that personal resources (Kim, 2017), illness severity (Killian et al., 2016) and positive meaning making (Cadell et al., 2014) may predict positive psychosocial responses (e.g. closer relationships, improved coping) in the context of parenting a child with LLC and LTC (Cadell et al., 2014; Ware & Raval, 2007). However, factors that might be liable to guide psychosocial support for parents (Williamson, 2018), or enhance their psychiatric health status are not examined routinely (Mantulak & Cadell, 2018). Hence, it is difficult to interpret any variation in emotional response from one parent to another (Veberne et al., 2017). Understanding the core life experiences that are inherently therapeutic for parents of children with LLC and LTC remains a key research priority (NICE, 2016), especially in the context of the review study (chapter 1), which identified a prevalence of distress within this population. A better knowledge of the experiential phenomena that lead to positive emotion and wellbeing in this population is required, before clinical support in PPC can be fully optimised (Huot & Fitzpatrick, 2018).

The experiential activities that have psychologically positive and therapeutic implications in parent caregivers need to be delineated. Given the lack of research in this area, it may be important to draw on ideas from the narrative therapy (NT) field. Following the original work of Bruner (1991) contemporary narrative theorists (Schenker et al., 2015) have consistently suggested that individuals encountering highly traumatic life events are able to find wide ranging social and emotional benefits from the act of talking or writing about their life experiences (Pennebaker, 2000). By drawing principally on research in the fields of PTSD (Robjant & Fazel, 2010) and narrative psychotherapy (Neimeyer, 2004) Pennebaker, J. (1997, 2018) has argued that narrative approaches using a storytelling framework may contribute to the development of emotional health and foster enhanced psychological adaptation following trauma (Pennebaker, 2000). The terms ‘narrative’ and ‘story’ are typically used interchangeably by Pennebaker (1993) and colleagues (Ryan, 2007; Elmes & Barry, 2017), to describe ‘thematically sequenced accounts’ which ‘convey meaning’ from one individual to another through written or spoken language (Elmes & Barry, 2017, p.4).

Confiding or account making through telling one’s story of adversity is considered especially helpful for facilitating psychosocial wellbeing and establishing a framework for managing traumatic upheaval in the context of a highly stressful life event (Pennebaker, 1997; Neimeyer, 2004). According to Pennebaker’s (1997, 2000) *emotional disclosure theory* stories can be constructed to integrate and transcend personally difficult life experiences. Typically, storytelling in relation to a painful or upsetting event is thought to act as a powerful therapeutic agent, which leads to diverse improvements in emotional health and wellbeing (e.g. increased life satisfaction, closer relationships, improved confidence, reduced depression, reduced distress, fewer physician visits) in comparison to control topics (Pennebaker, 1997, 2000; Pennebaker & Stone, 2004; Frattaroli, 2006). The underlying premise of the theory is that recounting traumatic life narratives through a story is a therapeutic experience (Graybeal, Sexton, & Pennebaker, 2002), as it allows the individual to (a) reconsider negative thoughts about the event (b) integrate new information into the trauma narrative (c) process difficult emotion and (d) find benefit in adversity (Pennebaker, 2000; Harber & Pennebaker, 1992). Particularly where individuals are able to receive validation, or social support by telling their telling their story in a safe environment, this in turn is thought to aid cognitive processing and provide a context for positive emotional change (Davison, Pennebaker, Dickerson, 2000; Pennebaker & O’Heeron, 1984). Although pervasive disorganised narratives may emerge immediately following a traumatic incident, Pennebaker (2000) suggests that stories promote integration and transcendence of the trauma event. They help the client to develop a coherent, meaningful explanation of the trauma they encountered, which may ultimately have an ameliorative impact on the sense of grief, pain and anxiety that is frequently experienced in response to a major life crisis and loss (Pennebaker, 1997; Neimeyer, 2004).

At a practical level, evidence to corroborate Pennebaker’s theory (1997, 2000) has come from narrative interventions that make use of storytelling (Neuner et al., 2008). Creative therapies that involve written or oral discussion of trauma have been shown to have significant positive effects on emotional health and wellbeing among emergency service workers (Alghamdi, Hunt, & Thomas, 2015), university students (Pennebaker, 1997), war veterans (Thiessen, 2013) and rape victims (Foa, Molnar, & Cashman, 1995). As the majority of these interventions involve laboratory based expressive writing (for 15-20 minutes; Pennebaker, 1993) or structured storytelling in a group format they are typically examined within a positivist framework (Schenker et al., 2015). The frequent use of quantitative surveys, alongside meta-analytic research implies a methodological commitment to outcome-based measurement (Travagin, Margola, & Rebson, 2015). Inductive approaches, such as qualitative interviewing are routinely omitted in favour of a quantitative paradigm that assesses causal relationships between isolated variables, using conventional statistical techniques (Yilmaz, 2013). This objectivist method of inquiry precludes a more fine-grained analysis involving the idiosyncratic experiences of each storyteller (Smith, 2011), and means we seldom learn about the individualised aspects of the storytelling experience (Pennebaker, 2000), or what it is like for a person to tell their story in their own words (DiFulvio et al., 2016).

Opportunities to understand the way storytelling is individually experienced and socially understood are scarce within the extant literature. Thus, a further ideographically orientated qualitative analysis of this phenomenon is timely. Indeed, until such analyses are conducted it will be difficult to understand the full complement of health benefits that storytelling is able to afford. Moving beyond outcome measurement and meta-analytic research has been identified as a recent goal in the NT literature (Nurser, Rushworth, Shakespeare, & Williams, 2018), which may help to further elucidate Pennebaker's theory (2017) and the wider experiences associated with a storytelling disclosure.

To date, explorations in NT have primarily been limited to the studying storytelling from a non-paediatric perspective. Thus, the theoretical ideas presented in this chapter are yet to be examined in parents of children with LLC and LTC. Whilst Pennebaker (2004) and his associates (Romanoff &

Thompson, 2006) have suggested the possibility of investigating storytelling in palliative care populations, studies in the healthcare field have tended to critically evaluate storytelling in somatically ill adults (Morina et al., 2012), interdisciplinary oncology professionals, or bereaved caregivers (St-Louis & Bourjolly, 2018; Roepke et al., 2018). There is subsequently an ongoing need to examine the potential impact storytelling has in parents of children with severe illness (Wilson & Chando, 2015). Indeed, the high rates of distress within this population indicate that storytelling may well also be valuable to parents or care providers affected by ongoing paediatric illness concerns (Cadell et al., 2012). Parents of children with LLCs and LTCs often encounter a unique series of losses and disease-specific traumas that occur over the illness life course (Steele, 2016). Adversity may be prolonged especially in cases where medical care is able to increase the child's life expectancy. Considerable long-term difficulties exist for many parents because even if 'the child's terminal prognosis is known… the exact length of time until death is not' (Cadell et al., 2014, p.130). Traumatic exposure is commonly acknowledged in this population (Cadell et al., 2012), although the psychological theory of Pennebaker (2000) requires further interpretation and elaboration with regards to parents of children with LLC and LTC. Research is needed to explore the pertinence of Pennebaker's theory in relation to these parents, given the extended nature of the stresses they describe (Veberne et al., 2017), and the potential storytelling has to improve their quality of life (Neimeyer, Pennebaker, & Van Dyke, 2009).

Paediatric Palliative Care

Telling one's story of trauma involves describing the event or situation in words (Ewens, Hendricks & Sundin, 2017). As this process can aid positive psychological change and transformation, palliative care practitioners have increasingly sought to make sense of illness and death through storytelling approaches. Oral history taking has a long history and tradition in UK PPC settings, although interest in hearing patient and family stories has increased over the last fifty years due to growth of holistic, family centered models of care (Bingley et al., 2006). The aspiration from Together For Short Lives, a leading UK charity for LLC and LTC is now that every parent of a life-limited child will receive care that takes into account their personal illness story (TFSL, 2018), thereby ensuring the provision of effective emotional and psychological support for these individuals from diagnosis into bereavement (TFSL, 2013).

Recommendations from the UK Department of Health (DoH) therefore include the suggestion that parents of these children should be regularly offered the opportunity to share their stories and experiences with other families caring for children with similar illness conditions (DoH, 2013). Listening to the parents story is considered essential for ensuring the delivery of competent, high-quality professional care in modern PPC services (Davies, Davis, & Sibert, 2003). Within the children's hospice sector narrative therapies are often considered a core aspect of the family care plan, and parents are frequently encouraged to develop their skills in storytelling through public advocacy roles (NHS England, 2015). Parents have recently become involved in online medical campaigns to raise awareness about LLCs/LTCs (GOSH, 2018), or otherwise shared their story of caregiving in an educational capacity through the news media (Caddel, Kennedy, & Hemsworth, 2012). Public speaking, as well as support groups have expanded rapidly in response to a perceived lack of knowledge about LLC or LTC illness (Rafferty & Sullivan, 2017; Contact, 2018). As a result parents now have more opportunities than ever before to raise concerns or share information regarding care provision through the medium of storytelling (Avieli & Band-Winterstein, 2017), both within children's services and via their own grassroots efforts (TFSL, 2018).

While practices involving the disclosure of an illness story are increasingly popular, little is known about the experiences of parents who have told their story of caring for a child with LLC or LTC. At present there is a dearth of research exploring the lived experience of parent caregivers as storytellers. Empirical studies have not investigated the impact that telling one's story may have upon mothers and fathers who are supporting a child with serious illness needs. Due to the growing application of storytelling in PPC services, a detailed examination of parents' experiences in this area is timely, and needed before we can adequately appreciate what it is like for these individuals to relate their stories of caring for children with LLCs and LTCs at service-led groups or events. The narrative theory proposed by Pennebaker (1997, 2000) also proposes that storytelling is likely to be perceived as a psychologically positive and therapeutic experience, although this finding has not yet been observed in a palliative context with parents of children with LLC and LTC.

Chapter three of this thesis outlines a novel empirical study that seeks to collectively address these issues. The study explores parents' experiences of telling their story of caring for a child with a LLTC or LTC. Results from this study will seek to inform evidence-based clinical practice in PPC services, and assess the potential implications of storytelling for parents in future paediatric palliative settings.

3.

Empirical Research Paper (ERP)

A qualitative IPA study examining the experience of storytelling among parents of children with life-limiting conditions (LLC) and life-threatening conditions (LTC) in a UK paediatric palliative care context

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Abstract

Objective: A paucity of literature exists concerning parents of children with life-limiting conditions (LLCs) and life-threatening conditions (LTCs). This study therefore aimed to explore parents' experiences of telling their story of caring for a child with a LLC or LTC, both a) in a UK children's hospice context and b) in the community.

Design: This study adopted a qualitative research design.

Method: Face-to-face, semi-structured interviews were conducted with eight parents, recruited from a UK children's hospice charity. All parents were actively caring for one child with a LLC or LTC at the time of interview. Interviews were subjected to Interpretative Phenomenological Analysis (IPA).

Results: From the IPA five superordinate themes emerged: a)'bonding with other parents through storytelling' b) 'therapeutic storytelling to a hospice professional'c) 'storytelling as an educational tool' d) 'fear of others reactions to the story' e) 'weariness through repetition of the story'.

Conclusion: Themes reflected the understanding that storytelling is often a psychologically positive and therapeutic experience for parents. Parents predominantly benefited from telling their story in a children's hospice context, although the negative aspects of storytelling predominated in the community. Further exploration of storytelling in parents of children with LLC and LTC is warranted.

Keywords: Storytelling; parent; life-limiting condition; life-threatening condition; hospice; IPA.

Introduction

Life-limiting conditions (LLCs) are illnesses where there are no reasonable prospects of receiving curative treatment and where premature death is considered likely or inevitable (Together for Short Lives; TFSL, 2013). Life-threatening conditions (LTCs) are illnesses where curative treatment options may be feasible but can fail (TSFL, 2013). In the UK, 40,000 children are diagnosed with an LLC or LTC (Popejoy, Pollock, Almack, Manning, Johnston, 2017). Rates of diagnosis are also increasing (Plunkett & Parslow, 2016). Thus, a growing number of parents are expected to support their children in the community, despite the significant emotional and practical challenges this presents (Bluebond Langner et al., 2014).

This situation poses difficulties at an individual and societal level (Ware & Raval, 2007), as family carers, especially parents, face overall responsibility for managing the child's care (Lotz, Daxer, Jox, Borasio, & Fuhrer, 2017). High levels of associated isolation, distress and psychopathology result from the demands placed on these parents (Collins et al., 2016). Parents of children who have been diagnosed with a LLC or LTC experience a variety of traumatic life situations, in which the uncertainty and impending threat of the child's death leads to an increased rate of mental ill-health (Bally et al., 2018). Following their child's admission to paediatric intensive care, it is estimated that 84% of parents exhibit posttraumatic stress symptoms, while 10%-21% develop post-traumatic stress disorder (Nelson & Gold, 2012).

However, despite significant emotional turmoil and impairment, healthcare professionals note the relative absence of published psychological approaches in paediatric palliative care (PPC), that are designed to ease parental burden and distress (Manguy et al., 2018). With the possible exception of respite (Ling, 2012), few therapeutic modalities have been studied in detail. Therefore, psychosocial support preferences among parents of sick children remain unclear in both LLC and LTC populations (NICE, 2016).

Storytelling

One particular therapeutic phenomenon that requires further study in this population is storytelling (Akard et al., 2015). For the purpose of this study, the terms 'storytelling', and 'telling one's story' are used to describe paediatric illness-related discourses that pertain to caring for a child with LLC or LTC (Bingley, Thomas, Brown, Reeve, & Payne, 2008). In PPC, illness stories are thought to have a major therapeutic impact by allowing individuals to communicate understanding, make sense of their experiences (Wangmo et al, 2017), find meaning (Laing, Moules, Estefan & Lang, 2017) or promote feelings of closeness to others in a supportive healthcare setting (Keesee, Currier, & Neimeyer, 2007). Stories provide a clear way for individuals affected by palliative illness concerns to create order and continuity from the chaotic disruption of critical illness and repeated hospitalisation experiences (Manning, Hemingway, & Hedsell, 2017), while also serving to legitimise ones difficulties at times of uncertainty (Bosticco & Thompson, 2005) and loss (Neimeyer, 2006).

In the UK, stories have long been considered to have therapeutic value (Gunaratnam & Oliviere, 2009). Storytelling has been used in various guises within the palliative care sector since the early 1980's (Laing et al, 2017). More recently, however, in UK-based PPC settings, there has been a move to incorporate storytelling into everyday clinical practice (Wilson, Hutson, & Wyatt, 2015). Parents in contemporary hospice services are often encouraged to relate a single, defined and comprehensive story of caring for their ill child (Gunaratnam & Oliviere, 2009). These stories have been recorded both privately in a therapeutic sense (Neimeyer, 2012), as well as publicly in the social media domain (Somanadhan & Larkin, 2016). Storytelling engagement with parents inside the children's hospice sector has often involved interactive dialogue with a nursing staff member or counselor (Wilkinson, Croy, King, & Barnes, 2007), while online forums, technology based interventions, fundraising events and support groups have also led to more public forms of disclosure (Martin et al., 2018; Rafferty & Sullivan, 2017). Hospices therefore frequently seek to help parents order and integrate their experiences as caregivers through storytelling (Kirk & McManus, 2012).

The act of storytelling is central to the ethos of modern UK hospice services (Gunaratnam & Oliviere, 2009). Storytelling forms part of the dominant etiquette and culture within the hospice environment. Due to the importance of storytelling within the hospice clear examples of parents stories are provided online by organisations including East Anglias Children's Hospices (https://www.each.org.uk/why-we-do-it/hear-from-families) Children's Hospice South West (https://www.chsw.org.uk/stories/family-stories) and TFSL (https://www.togetherforshortlives.org.uk/get-support/your-childs-care/family-stories/). The stories parents tell are typically centered on their practical and emotional struggles when caring for their ill child. Whilst these stories are often told formally to an audience within the hospice, the popularity of storytelling has also led some parents to repeat their story in the community as an educational tool to raise awareness regarding paediatric illness conditions.

Despite the widespread use of storytelling in UK PPC, very little is known about the experience of storytelling. Overall, few empirical studies in PPC have explored this phenomenon (Barnato et al, 2017). Storytelling has been investigated in UK children with serious illnesses (Freeman, 1991) but not their parents, and it is recognisable that further family-orientated research is needed (Wilson et al, 2015). This study aimed to address this gap in the literature by exploring the phenomenon of storytelling. The parental experience of storytelling was therefore examined a) in a UK children's hospice context, for example at fundraising events, support groups, and b) in the community, for example in familial conversation, or via an online blog. Qualitatively exploring this phenomenon will help to provide insight into the act of storytelling, its implications for psychosocial wellbeing, and guide the delivery of parent-centered storytelling in future. The primary question in this study is, therefore, 'what are parents' experiences of telling their story of caring for a child with a LLC or LTC?'

Method

Design

This study adopted a qualitative research design using Interpretative Phenomenological Analysis (IPA). The IPA framework and guidelines of Smith, Flowers & Larkin (2009) were followed throughout. As a method, the aim of IPA is to thoroughly and systematically examine a person's involvement in a particular experience, event, activity, or process (Smith et al., 2009). IPA is considered appropriate for exploring the area under investigation because it seeks to gather detailed, insightful accounts from individual study participants and explore how people make sense of their world (Larkin & Thompson, 2012). As IPA is primarily interested in the idiosyncratic nature of experience, it is well suited to the study of high-individualised encounters in a palliative care setting. Indeed, IPA is predominantly used to explore life-changing illness phenomena in-detail (Smith, 2011), thereby making it suitable for an experiential hospice-based study involving parents of children with LLC and LTC (Smith et al., 2006). Due to the interpretative nature of IPA, the research team approached this project from a contextualist standpoint, which assumes reality is both subjective (MacFarlane. 2007), and socially negotiated (Braun & Clarke, 2013).

Organisational context

Recruitment for this study is purposive (Smith & Osbon, 2007). The participants were a homogenous group of parents recruited from a single hospice organisation. Parents were recruited from a registered children's hospice charity[4] in the UK, supporting both children with LLC/LTC and their families. The charity has ownership of three separate children's hospice sites. The ethos within the hospice charity is informed by a narrative therapy approach. Storytelling therefore constitutes a core activity within the hospice. The charity employs a wide range of multidisciplinary clinical staff (e.g. nursing staff, counsellors) and encourages storytelling as a method of self-

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[4] The actual name for is omitted for anonymisation purposes.

expression for parents. In order to build connectedness with service users and staff, parents' stories are typically disclosed formally at hospice support groups and events, as well as through counselling sessions within the hospice. Parents also tell their story more broadly through external educational initiatives, medical appointments, conversations with laypersons, and through hospice-led fundraising initiatives in the community. The stories parents' tell are extended in duration and guided by particular content as they typically convey the parents practical and emotional challenges of caring for their ill child.

Inclusion/Exclusion Criteria

Selection and recruitment processes for the study were constructed collaboratively within input from parent service-users/staff at the children's hospice charity (Appendix F), and were also reviewed by parents at a second national PPC organisation, prior to implementation (Appendix G):

Inclusion Criteria

- A parent with lived experience caring for a child (aged 19 or under) with a diagnosed LLC or LTC.
- The parent is the child's biological parent, foster parent, or legal guardian.
- The parent previously discussed their story of caring for a child with a LLC or LTC at a group or event led by the children's hospice charity and/or discussed this story in everyday life/more generally in conversation with other people outside the children's hospice charity.
- The parent is aged ≥18, and able to speak conversational English without the use of an interpreter.

Exclusion Criteria

- A parent with a child placed on the end-of-life care pathway by the children's hospice charity. Staff at the hospice charity recommended this exclusion criterion, as death of the child is imminent and the narratives of these parents were considered likely to be dominated by anticipatory grief (Smith et al., 2015).

Recruitment

Participants meeting the inclusion criteria were recruited from each of the three hospice sites between January-May 2018. To maximise recruitment, staff provided parents with recruitment flyers and information sheets at relevant hospice led groups and events. Parents were also given the option to contact the researcher about participation via the bi-monthly service newsletter (appendix H), through an online advert (appendix I) and through flyers (appendix J) placed in hospice reception areas. The primary author (T.M) checked eligibility via telephone or email, then met in-person with the parent to obtain written informed consent prior to participation. Details of the participants were anonymised following data collection, and stored confidentially at the University of East Anglia (UEA). This study received ethical approval from the Faculty of Medicine and Health Sciences at UEA (Appendix K) and the Board of Clinical Governance at the children's hospice charity (Appendix L).

Participants

A total of 8 parents participated in the study (table 1). Participants were 6 mothers, 2 fathers, and were Caucasian (n=7) or Asian (n=1) aged 25-54 years (n=8). They were either full-time carers (n=4) or in employment (n=4). All parents were actively caring for their ill child at the time of interview. The primary LLC-LTC diagnoses of children (n=7[5]) were heterogeneous and included apert syndrome (n=1); lissencephaly (n=1); cerebral palsy (n=2); mitochondrial myopathy (n=1); metachromatic leukodystrophy (n=1) and epileptic encephalopathy (n=1).

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[5] One couple (two parents of the same child) participated in separate interviews.

Table 1. Participant characteristics

Participant (P)[i]	Gender	Age	Ethnicity	Occupational status
'Jane'	female	25-34	white british	part time employment
'Deborah'	female	25-34	white british	full time carer
'Christine'	female	35-44	white british	full time carer
'Rita'	female	35-44	white british	full time carer
'Max'	male	45-54	white british	employed full time
'David'	male	35-44	white british	employed full time
'Emma'	female	45-54	white british	full time carer
'Hannah'	female	35-44	Asian	employed full time

[i] Pseudonyms are used for all names. The sample included parents from the three hospice sites.

Data Collection

In-depth, audio-recorded semi-structured interviews were conducted individually with each participant in a quiet, comfortable location at their home address, or workplace (Smith, 2004). As per recommendations from Smith et al. (2009), each interview lasted 45-79 minutes to allow for a deep and rich exploration of the participants lived experiences' (Pietkiewicz & Smith, 2014). Interviews were conducted exclusively by the first author (T.M). Participants were asked a series of probing, open-ended questions from an interview topic guide (Smith et al., 2009). The interview topic guide contained six non-directive questions that were designed to elicit detailed information about the target phenomenon (e.g. 'what sort of things of are you aware of when telling your story at a group or event run by the hospice?). In order to authentically enter the lifeworld of the participant, the first author used the topic guide flexibly (Hunt & Smith, 2004). Any interesting or unexpected details were discussed further to ensure the nuances of each experience were captured thoroughly in the participants' own words (Smith, Spiers, Simpson & Nicholls, 2017).

Analysis

Completed interviews were transcribed verbatim by the first author and subjected to IPA (Smith et al., 2009) in NVivo 11 (QSR International, 2017). Transcripts were examined individually and re-read several times, prior to line-by-line commenting. Exploratory comments were typed in the wide margins of the transcript to note points of verbal, descriptive and conceptual interest (Smith et al., 2009). The comments were then further analysed to develop themes. Interconnections between each theme were searched for and any interrelated themes were merged to create superordinate themes (Smith et al., 2009). Transcripts were continually re-read to ensure each theme retained its original link to the data (Nizza, Britton, & Smith, 2016). Once all the transcripts had been analysed separately using this procedure, the first author developed a comprehensive, consolidated list, containing all the superordinate themes. The superordinate themes were then reviewed, reconfigured, subjected to further scrutiny by the second author, and rearranged, until a final list emerged. The consolidated superordinate themes were then presented in a narrative form within the results section of the present article.

Rigour

Procedures for increasing the rigour of this study were adhered to throughout (Yardley, 2000). The first author kept a reflective diary throughout the research project, in order to ensure continued monitoring of any personal assumptions or attitudes that could unduly influence the data collection and analysis process (Vicary, Young, & Hicks, 2017). Regular supervision (Ware & Raval, 2007), NVivo training, and involvement in an IPA discussion group (http://www.ipa.bbk.ac.uk/discussion-group) were further analytical strategies employed by the first author to minimise bias (Draper & Swift, 2010). The first author attended the hospice charity regularly on an informal basis during the course of the study. This allowed for interpretations that were informed by a more enriched socio-cultural understanding of the participants' lifeworld (Yardley, 2000). Input from parent service users attending the hospice sites also ensured the topic guide questions were developed collaboratively and were not simply informed by the subjectivities of an individual researcher (Rodham, Fox, & Doran, 2015),. Participants consenting to member validation (n=4) were sent a copy of their verbatim transcript and asked to confirm that it reflected their experiences accurately. In all cases, participants stated that their transcripts were accurate (McCann et al., 2012). Emergent themes were identified within the transcripts by the first author, and the second author (K.M.) then checked each interpretation to ensure accuracy. Any disagreements were resolved through further collaborative discussion.

Position

IPA is a reflexive method and therefore it is important for the principal researcher to make clear his own position as it relates to PPC (Dalby, Sperlinger, & Boddington, 2012). The first author acknowledges that he is not a parent of a child with a LLC or LTC. However, he moved closer to an ‘emic’, insider position during this study (Gil, 2015) by informally attending hospice events (e.g. parent support groups), working in a General Paediatric Health Service and reading widely around the subject of PPC.

Results

Through the IPA five superordinate themes were identified. Each theme represented a different core aspect of the storytelling experience. The superordinate themes reflected experiences of storytelling both within a hospice context (for example at hospice fundraising events, support groups or in counseling sessions) and more generally in the community. The identified superordinate themes were a)‘bonding with other parents through storytelling ’ b) ‘therapeutic storytelling to a hospice professional’ c) ‘storytelling as an educational tool’ d) ‘fear of others reactions to the story’ e) ‘weariness through repetition of the story’. Frequency counts, indicating the overall number of parents identifying with each superordinate theme, are represented in title brackets.

Bonding with other parents through storytelling (8/8)

The theme 'bonding with other parents through storytelling' reflected parents' feelings of closeness to their peers when telling their story in a hospice setting. Mothers and fathers described a belief that telling the story at support groups in the hospice helped to cultivate new relationships, particularly with other parents that were caring for children with similar illness conditions. They experienced a sense of connection to these parents, who listened attentively and could sympathise with their situation:

'I go to a monthly hospice group where other parents are…they understand what you mean, when you say you've had a bad few weeks, a gazillion hospital appointments on top of maybe him not sleeping properly…they understand where you are coming from. They can sympathise, rather than just empathise, unlike a friend who doesn't come from that world'.-'Christine'.

Mothers and fathers felt that they were able to bond emotionally with one another over similar stories ('Hannah', 'David'). By exchanging their worries & fears about LLC & LTC through storytelling parents learnt that they were metaphorically 'in the same boat' as some of the other families, who also attended the hospice ('Max', 'Christine'). A sense of togetherness emerged, in which certain parents felt that telling their story had allowed them to cultivate a close network of friends, confidantes and allies, who could be called on to provide emotional support when needed:

'When you are having problems they can solve it. So it's always good to be able to bounce ideas off each other and try and help each other, or just have a good whinge, or a good cry, or a good laugh'. 'We felt kind of lonely to begin with, and to be able to talk about our story and get it out there, actually, we weren't alone'. –'Rita'.

Parents who imparted their story often felt less isolated. When they were able to share their story & interact with other families they experienced a feeling of kinship ('Jane', 'Rita', 'Hannah'). There was a sense that other parents of children at hospices & other PPC services, had a unique, 'shared history', or acted as a form of 'extended' family ('Deborah'), which made it easier to tell them the story. Parents attending a hospice subsequently experienced a feeling of trust towards one another, which one interviewee touched upon:

'Well (when you tell the story) you get the feeling that other parents 'get it', especially at the hospice, but we don't have that many opportunities to meet with many other parents with kids with additional needs around here. So, you, you sort of get a level of empathy that you don't get elsewhere'.- 'Jane'.

This connection informed the willingness of some parents to seek practical advice from other families. Tips and guidance were frequently sought, particularly during the early stages of the illness. One parent told her story to other parents in a hospice group, in order to inform the management of his particular mobility problems:

'We'd gone from everything being, fine, to within, very quickly, having lots of issues with mobility and needing to sort a lot of stuff out, very quickly... there were quite a few people (at the group) who are in the situation where their children have been erm disabled from birth, so they've had y'know years of experience and so...people could say, oh yes, what we've got, this works. I went around to somebody's house to see their bathroom adaptation, and it was those sorts of opportunities that I found really good, because we just didn't have any other means. You can't go into a shop down the road'. – 'Emma'.

Parents did however find it more difficult to share their story with certain families. If the child's diagnosis differed ('Jane'), or the care needs diverged substantially it could feel harder to impart the story ('Max', 'Rita'). Thus, similarities were needed to ensure that storytelling functioned as a bonding experience for parents ('Christine'). The context of storytelling within the hospice seemed to be particularly important, as parents reported that this was the principal environment where they could tell their story and feel close to individuals who shared their difficulties as caregivers.

Therapeutic storytelling to a hospice professional (8/8)

In the theme 'therapeutic storytelling storytelling to a hospice professional' parents reported a therapeutic feeling of being accepted and understood when telling their story to a compassionate professional in the hospice. Often, parents encountered this experience when telling the story to a counselor within a hospice (‘Jane’, ‘Deborah’, ‘Christine’, ‘David’, ‘Hannah’). The counselor reflected on their stories of adversity and helped to validate the difficulties they faced. A sense of acknowledgment pervaded one mothers experience when disclosing the negative aspects of her story to a counsellor:

‘But I think she just, she didn’t try and like ‘there there’ it will be ok approach. She was realistic. It is rubbish. But there are things that can make it less rubbish…she listens but then gives you some um, pragmatic things to think about and act on’.-‘Christine’.

Parents appreciated the experience of relating the story to a compassionate, non-judgmental figure. There was a sense that telling the story to a counsellor freed parents from some of the usual expectations and prejudices that existed in the outside world. In this context some parents felt a sense of security (‘Jane’, ‘Christine’). A warm, private and person-centered ethos to counselling sessions meant that parents often felt less vulnerability when telling their story as part of a therapeutic dialogue:

Interviewer: ‘you mentioned telling the story to a counsellor or psychologist…what has that been like?’

Participant: ‘absolutely brilliant, fantastic, I couldn’t see ever not doing it if that makes sense. I found it so helpful just to have…a space, every 2 months to just talk about what’s been going on… it’s just having a safe space to talk’.-‘David’.

Experiencing this compassion also helped to alleviate distress. Telling the story to a counsellor felt cathartic. It allowed parents to begin processing their emotions, and openly express the anguish they were experiencing in a helpful way:

‘Nearly every session with the counsellor I cry a little bit, because there is something, and I think, the counsellors, they are professionals, so they are quite good…they understand the situation in this family…you will tell them and after…you feel better. Or at least during that hour you will feel better’.-‘Hannah’.

Parents expressed feelings of warmth toward other hospice staff and valued their empathy (‘Rita’, ‘Max’, ‘Emma’). A mother and father of the same child both felt that telling their story to compassionate and understanding staff member in the hospice constituted a therapeutic experience:

‘*I guess it was the first time we’d told anyone the story of the past 5 months…the staff member was really understanding’.-‘Max’.*

‘They (staff) were kind of like very supportive and you know managed to calm us down…and say this is what we can offer you’. ‘It’s also helped. It’s therapy, if you like, getting to talk about it’.-‘Rita’.

Through storytelling, parents encountered a therapeutic dialogue where they felt accepted and understood. Hospice staff and counselors were considered especially skilled in their ability to cultivate these feelings and mitigate distress through their interactions (Jane, Christine). Subsequently, the hospice professional was valued among parents for helping to facilitate positive therapeutic storytelling experiences.

Storytelling as an educational tool (7/8)

Within the theme ‘storytelling as an educational tool’, parents indicated that they were able to derive a sense of meaning and purpose from telling the story to educate others. New life possibilities were identified as parents recognised that their story could be used to educate members of the public about LLC and LTC (‘Jane’, ‘Max’). Parents found that they were frequently able to educate others by telling the story at fundraising events led by the hospice charity (‘Rita’, ‘Max’, ‘Emma’), or, less commonly, at local gatherings in the community (‘Deborah’, ‘Christine’). Parents felt that they were able to take on an important societal role as they helped to shape public attitudes towards children with paediatric LLC/LTC through the disclosure of their story:

> *'I think that helps, to tell the story as well because people have watched and said 'oh we never realised you had to go through this and 'Janet'*[6] *has to go through that' and that's been a really good way to explain to people in layman's terms without trying to go too far into detail… it was really good to you know get that awareness out there'.-'Max'.*

One mother told her story at a local church. She valued this opportunity to give people an insight into her son's condition. Through telling the story, she also noticed how people's attitudes toward illness were able to change, and how her actions had a positive impact upon the audience:

> *'Recently I did a training session for the church…we got people blindfolded and with ear defenders, because our child is deaf and blind. We did 2 different exercises, to get people to think what it must be like for someone like that. We just did a simple game. One of the games we did, we blindfolded people and different people had to go and say hello to them. Seeing people engage with the activities and things dawning on them, just how awful it is… or 'oh' it's much better to do things like that'.-'Deborah'.*

A number of parents interviewed began telling their story educationally in a fundraising context. At charity events, parents indicated that they were able to enlighten others ('Emma'), as well as raise large sums of money ('Max'). One of the parents interviewed reflected on the sense of empowerment she derived when telling her story educationally as part of a hospice event, which aimed to obtain additional funding for the service:

> *'At that event, because of telling our story with other parents, we raised a lot of money that night…we were being bombarded: 'if there's anything we can do, give us a shout.' A couple of audience members were like…what can we do to help? I said put it out there. Just put it out there that the fundraising appeal is so important to other families, because obviously the bigger it is the more families that can help… I mean, that was amazing'.-'Rita'.*

Parents also benefited emotionally from telling their story to educate others. They encountered a sense of strength and wellbeing through storytelling, as expressed through increased confidence ('Deborah'), pride ('Max', 'Emma'), hope ('Jane'), faith in others ('Rita') and feelings of personal liberation ('Christine'). One parent believed that telling the story through research gave

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[6] Pseudonyms are also used (e.g. 'Janet') for all child names.

her narrative wider influence. This seemed to result in a sense of self-efficacy and empowerment, as there was a belief the story could be used to help other parents:

Interviewer: 'And how does it feel I guess, to tell your story to me in this situation?'

Participant: 'I'm very happy to tell you. Because the reason for me to take part...I'm hoping my experience can be used in the research and then hope, that when the research is finished…that maybe it will be helpful for other parents'.-'Hannah'.

Storytelling in an educational context provided parents with a sense of meaning and purpose. There was a belief that they were enhancing education and increasing public knowledge through storytelling ('Hannah', 'Deborah'). In telling the story in this way parents also experienced positive emotional changes themselves.

Fear of others reactions to the story (8/8)

The theme, 'fear of others reactions to the story', relates to way that parents worried about the reactions of friends, family members, and strangers to the story. This theme did not arise at a hospice with other parents of seriously ill children, but appeared to be common in other everyday situations beyond the hospice. In a number of cases parents felt anxiety ('Jane', 'Deborah', 'Hannah'), as there was a sense that they could be subject to high levels of critical evaluation and scrutiny when talking about their child ('Jane', 'Deborah', 'Rita', 'Max', 'Emma', 'Hannah', 'David'). One parent intermittently felt uncomfortable sharing her story. She wanted to protect her family from the full story, but also expressed concern that family members might not respond empathically:

'So I just tell them (the story) but I don't go into detail. I don't want to worry them, but secondly maybe they wouldn't understand that much. Because like, when my daughter just started the seizures…I didn't tell them'.-'Hannah'.

Parents justifiably expressed concern about telling their story to members of the public, who did not typically have a well-informed understanding of paediatric illness conditions and their impact upon the family system. There was a sense that some parents were wary of discussing their child's illness with a stranger, who might hold stigmatising, or prejudicial attitudes towards themselves & their disabled child. One mother described how she carefully monitored the reactions of strangers when telling her story, before deciding how much more information to impart:

'So if there is a certain tone then…I decide that's it, I'm not going to bother. I put my wall up and that's where it will stay. And although I'm quite happy to talk about it I'm very like, do I trust you enough to want to talk to you about it?...So it does depend on the situation and how other people are'.-'Rita'.

A number of parents explained how they encountered difficulties when telling their story. Individuals were occasionally met with silence ('Jane'). Others were asked inappropriate questions via friends ('Deborah'), or about aspects of the story that they would rather keep private ('Emma'). This fed into the concern that members of the public were often likely to make inappropriate judgments about the reality of parenting a child with complex needs ('Christine'):

'There's people who… I am a bit uncomfortable with sharing certain information with…if you look and think oh…they didn't quite get what I was trying to say, nobody else really notices, but its just a bit erm, you feel like it matters more than it does'.-'Emma'.

'Well in regards to family, we've kind of spoken to them but erm, even after 6 years, I don't think my parents fully get what we have to go through on a daily basis'.-'Max'.

Although parents did continue to tell their stories, they were vigilant in doing so. A mother acknowledged that the fact that although she regularly told her son's story online, she remained cautious in her approach, and sought to limit its remit to minimise the probability of receiving a hurtful response:

'On my blog I have a privacy setting so people have to know about it to find it, because I think, I worry a lot. I know there are people out there who would think - that we shouldn't have had

him, that his life is not worth living, he's a drain on the system… You are very conscious about what other people think. It's, a bit, especially on the Internet you are more guarded'.- 'Deborah'.

Parents had difficulty disclosing in the community due to worries about the reactions of family members, strangers and friends. Parents were often concerned about negative reactions to the story where a lack of insight or empathy might be displayed. This led some parents to censor or leave out important aspects of their story, which perhaps also limited public knowledge of their difficulties.

Weariness through repetition of the story (7/8)

'Weariness through repetition of the story' reflected the dissatisfaction and fatigue parents could feel in their storytelling to statutory healthcare providers (HCPs), as they were often asked to repeat the story continuously. In statutory services, organisational factors (e.g. high staff turnover, time-limitations) affected the retention of the story & meant that storytelling became a monotonous, repetitive activity ('Deborah', 'Christine', 'Hannah'). Indeed, parents were frequently asked to retell the story at each hospital visit or medical appointment ('Rita', 'Max'). One father described the sense of disillusionment he experienced when he had to repeat details of the story on a hospital ward:

'But it's frustrating when you have to keep repeating it again and again...to the same people…So it is frustrating and I know other parents we have spoken to have said the same thing. They have to keep explaining it over and over. If she gets admitted to the ward, you have to go through the whole story every single time'.-'Max'.

Parents often felt distressed by these interactions ('Jane', 'Deborah', 'Christine', 'Rita', 'Max', 'David', 'Hannah'). They lamented being asked to tell the story 'again and again' ('David', 'Deborah') to a plethora of healthcare providers, particularly when the child's story could be accessed through their medical notes. A number of parents felt that this repetition reflected a professional failure to read the patient's file, which already contained the story.

One mother implied that certain HCPs lacked the initiative needed to carefully read the story and remember the details, thus requiring her to repeat it continuously:

'It feels like, knowing some of the doctors that I know, they haven't read the notes. They probably haven't read the notes, or read them as fully, and to me ...I find that really negligent'.- 'Jane'.

Another mother disclosed a sense of fatigue and frustration with the experience of storytelling. This encouraged her to consider the idea of no longer imparting the story:

'Everywhere you go, they will ask the same question...the health service, they should have every patients record in the system...why do they keep asking the same question?...You keep telling it, you keep telling it. And then every time you tell, you cry. So there is no need to keep telling it'.- 'Hannah'.

As parents seemed to feel dissatisfied by these experiences they began to take steps to address this issue. Writing out the story in full and giving a copy to each professional constituted one strategy that a number of parents had used to promote greater interest in the story and reduce the amount of retelling needed ('David', 'Jane'). One parent also chose to tell her story creatively using a technique known as 'beads of courage'. This aimed to capture the interest of professionals & promote recall by telling the story in a visual way:

'Basically there's a key, as it were, and certain medical procedures earn you certain types of beads, so every time our child goes to see one of his doctors he gets a blue bead, in hospital he gets a yellow bead, and you collect these beads, so I've got this string of beads, that tell his story in a visual way... I have taken them into hospital before and the doctors can see what he has gone through'.- 'Deborah'.

Parents' encountered dissatisfaction and fatigue as they repeated the story continuously to statutory HCPs. Subsequently parents used visual or written aids to address the issue of repetition. In this way storytelling could also constitute an evolving creative experience and was used adaptively in response to difficult encounters.

Discussion

This in-depth IPA study sought to examine the experiences of parents who have told their story of caring for a child with a LLC or LTC. Experiences of storytelling were explored a) within a UK children's hospice context b) and more generally in community settings. Although diverse experiences of storytelling were inevitably reported across each of the parents interviewed (n=8), five superordinate themes emerged from the data: a)'bonding with other parents through storytelling' b) 'therapeutic storytelling to a hospice professional' c) 'storytelling as an educational tool' d) 'fear of others reactions to the story' e) 'weariness through repetition of the story'.

Overall, the findings indicate that storytelling has the potential to be a psychologically positive and therapeutic experience (Charon, 2001), particularly within the context of a UK children's hospice. Through storytelling, parents developed feelings of closeness to their peers (bonding with other parents through storytelling') , felt understood and accepted ('therapeutic storytelling to a hospice professional'), and identified a sense of purpose through telling the story educationally ('storytelling as an educational tool'). The positive experiences predominantly arose within the hospice context, where parents and professionals were receptive to the story. However the negative aspects of storytelling were predominantly noted in everyday settings in the community. For example, parents also worried about the reactions of others ('fear of others reactions to the story'), and could feel dissatisfied when repeating the story to statutory HCPs ('weariness through repetition of the story').

To our knowledge, this is the first study to qualitatively highlight the therapeutic benefits associated with storytelling in a UK children's hospice context. Mothers and fathers (n=8) of children with a wide range of LLCs and LTCs participated and spoke of a cathartic dimension to the storytelling experience via a hospice, both when telling the story privately and in public, through counseling sessions, support groups, staff conversations and fundraising events in the service. In line with the contemporary ethos of UK hospice services the experience of hospice storytelling appeared to be particularly valuable in helping parents develop strong affective bonds and feel an affilitation with others (Dunbar, Carter, & Brown, 2018). Telling the story in this context promoted therapeutic relationships involving other parent service users and staff, providing the parent storyteller with a subsequent opportunity to receive practical and emotional support within the hospice environment. This is consistent with the understanding that hospices in the UK constitute flourishing communities, which typically facilitate peer interaction, promote belonging, and improve parental quality of life (Gosine & Travasso, 2018).

Results indicate a helpful quality and value to the phenomenon of storytelling in a UK hospice context (Gunaratnam & Olivere, 2009). The theme 'bonding with other parents through storytelling' reflected parents' feelings of relatedness to their peers. They cultivated emotionally close bonds & social relationships with other families through storytelling in hospice-led groups. Being the parent of a child with LLC and LTC constitutes a rare, highly marginalising experience (Bally et al., 2018). While previous studies have tended to focus on resulting sense of social isolation (Ware & Raval, 2007), or exclusion that permeates parents' lives (Collins et al., 2016), this study indicates that parental storytelling can act as a potential antidote to this problem, providing a feeling of 'connectedness' to others, and facilitating meaningful relationship development between families (Nicholas et al., 2009).

The hospice offered further opportunities for therapeutic dialogue. In 'therapeutic storytelling to a hospice professional' parents spoke of the warmth, security, acceptance and understanding they experienced through storytelling to hospice employees. Research in this area has demonstrated that a therapeutic dialogue is often cultivated when hospice staff take a individualised, responsive and person-centred approach to interpersonal interaction (Norton, 2018). Kirk & Pritchard (2012) suggest that parents typically value these therapeutic relationships and the non-judgmental stance offered by professionals in the hospice, particularly as parents often lack time and resources to cultivate such relationships in the outside world (Collins et al., 2016). Hence, it may be that storytelling in the hospice context can facilitate cathartic interactions with staff and mitigate the distress that commonly results from the demands placed on these parents (Verberne et al., 2017). Additionally, it may also be important for other non-hospice paediatric services (community teams, hospice wards, neonatal intensive care units) to consider replicating this aspect of the hospice ethos and apply therapeutic storytelling activities that involve both parents and staff.

Telling one's story also enabled parents to make sense of, and manage the traumatic aspects of their experience (Nurser, Rusworth, Shakespeare, & Williams, 2018). In the theme 'storytelling as an educational tool' parents were able to derive a sense of meaning and purpose from educating others. Storytelling in this context constituted an empowering experience (Graci, Watts, & Fivush, 2018) that allowed parents to thrive in the context of adversity. Parents recounted their traumatic life narrative in a way that solicited understanding of their difficulties and challenged the attitudes of others, which in turn, contributed to a sense of purpose and meaningful engagement in the community (Neimeyer, 2006). In keeping with the wider literature (Rafferty & Sullivan, 2017), parents often engaged in educational roles that sought to inform opinion and improve lay knowledge of issues relating to LLC and LTC through storytelling.

Telling the story entailed a risk that others would not always respond respectfully, or with appropriate level of understanding. The more problematic, negatively orientated aspects of storytelling were clarified within the theme 'fear of others reactions to the story'. In this theme parents worried about the views & perceptions of the listener, particularly when speaking to family members, strangers, or friends. A number of studies have reported similar concerns (Ware & Raval, 2007; Cadell, Kennedy, & Hemsworth, 2012). Parents of children with LLC/LTC often experience difficulty trusting laypersons. These individuals are often regarded as "outsiders" who have a limited comprehension their child's needs (Verberne et al, 2017). A dialogue of openness is difficult to cultivate, as concerns regarding potentially difficult conversations (Malcolm et al., 2011), or the expression of insensitive views abound (Rafferty & Sullivan, 2017). The prejudices parents envisage are potentially grounded in the reality that many LLCs/LTCs are rare (Hain & Devins, 2011), which in turn, curtails a more adequate public knowledge and appreciative understanding of PPC issues (Somandhan & Larkin, 2016). More research is however needed to explore the impact that public perceptions have in terms of complicating empathic dialogue, and limiting the openness with which a parent feels able to tell their story.

The theme 'weariness through repetition of the story' relates to the way that parents could also feel fatigue and dissatisfaction when repeating their story to statutory HCPs. Studies of dialogue between parents and paediatric HCPs have tended to focus on the communication skills of clinicians and their interpersonal styles (Zwaanswijk et al., 2007), finding a number of barriers (e.g. time restrictions, staff turnover, inadequate training) that may lead to a suboptimal interaction (Davies, Davis, & Sibert, 2003) and lower quality of care (Mechanic, 2001). Staff often lack the specialised time and expertise needed to support the care needs of parents of children with LTCs and LLCs (Kirk & Pritchard, 2010), which may contribute to less positive experiences of telling story in a paediatric consultation, and the need to repeat details of the story over time. Whilst parents expressed dissatisfaction with these encounters, they were also able to find creative ways to address repetition of the story through the use of written or artistic methods.

Strengths & limitations

This study exhibits a number of strengths. It is a novel project, and provides insight into storytelling in UK parents of children with LLC and LTC, a previously un-researched topic. Research in the field of PPC is often limited to the study of paediatric oncology (Orsey et al., 2017). Therefore, the inclusion of parents of children with genetically rare, non-malignant LLCs is considered a further strength. Questions of reflexivity were addressed through various systematic techniques (i.e. reflective diary, member validation, and research supervision), which enabled more effective 'bracketing' of any pre-understandings of the researcher regarding the analysis (Shinebourne, 2011).

Limitations of the study included self-selection bias (Robinson, 2014), and a reliance on the experiences of eight parents. Although IPA privileges small samples (Smith, 2011), the recruitment of eight parents from a single hospice charity in the UK limited the theoretical and cultural transferability of the findings. Few fathers were also recruited (n=2). Accordingly, this led to an overrepresentation of mothers (n=6) and precluded a more detailed understanding of the differences in experience between men and women within the sample.

Parents' experiences also varied widely with the respect to the event or group in which they told their story. It could be argued that the heterogeneity of this sample is problematic and goes against IPA's homogeneity principle (Smith et al., 2017). In IPA research, a narrow and homogenous sample is considered preferable (Shaw, Senior, Peele, Cooke, & Donnelly, 2009). Finally, findings were synthesised thematically using a qualitative IPA method. As such, the results are interpretative. The subjectivity of the findings limits our ability to infer more widely about the nature and quality of storytelling as an experience (Smith, 2011).

Research implications

Due to a lack of research, further studies are needed to explore the experience of storytelling for parents of children with LLC and LTC. In this study storytelling occurred both in public and in private. Other novel studies are needed to investigate storytelling in the contexts described, to assess its importance and impact upon the parent. In future, efforts should be made to explore parents' experiences of storytelling in other PPC services, across cultures and national contexts, with a more equally weighted gender sample of mothers and fathers. A study,could also be conducted to explore the diverse therapeutic and non-therapeutic impacts of storytelling.

Clinical implications

The themes identified within this study have a number of clear implications for clinical practice. Firstly, increased efforts could be made to promote bonding and relationship development between parents in children's hospices through storytelling in a group context. This could help parents of children with LLC and LTC receive practical and emotional support from their peers. The delivery of an illness-specific peer-support group may also be useful for parents in identifying common areas of concern and 'connecting' families of children with similar diagnoses (Martin et al, 2018).

Secondly, parents may benefit therapeutically from the opportunity to tell their story privately in the hospice with a counselor, or a staff member . An opportunity interact with a hospice professional may have value by allowing the story to be received in an open understanding way via an appropriately trained clinician. Thirdly, the sense of meaning and purpose that parents derived from storytelling in an educational capacity, indicates that parents may also benefit from further involvement in advocacy and educational work. Parents could offer examples of telling their story as part of an educational event to promote feelings of empowerment. However, in order to manage barriers that may prevent parents from storytelling in public, it may also be pertinent for professionals to address parental fears and anxieties concerning others reactions.

Finally, medical professionals should seek to ensure adequate retention of the story and reduce its repetition among parents (Davies et al., 2003). In the context of organisational pressures, parents of children with LLC and LTC could use creative written or artistic methods in an effort to capture the interest of HCP's and reduce verbal retelling of the story.

Conclusion

This IPA study provides a rare insight into the lived experiences of parents who have told their story of caring for a child with a LLC and LTC. Experiences of storytelling were explored both a) within the context of a UK children's hospice and b) in the community. Overall, a number of the themes highlight the psychologically helpful nature of storytelling in the context of a children's hospice. Although parents benefited from sharing their story publicly in an attempt to inform and educate others, negative aspects of storytelling predominated outside the hospice. The results from this study have important implications for clinical research and practice, as they suggest that storytelling can offer meaningful, empowering and cathartic experiences for parents.

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4.

Extended Methodology

The extended methodology expands on the methodology outlined in Chapter 3. It includes a further discussion of IPA and its theoretical origins. The IPA sample and interview procedure are also further described. Finally, a more detailed step-by-step outline of the analysis is given, along with the steps undertaken to ensure rigour (Yardley, 2000).

IPA

In the empirical research paper (Chapter 3), a qualitative methodology was employed. An idiographic, qualitative framework had the advantage of thoroughly and systematically exploring the lived perspectives and experiences of each participant in detail (Flick, 2014). The application of this approach helped to authentically represent parental voices within the study, and allowed for a deep and nuanced understanding of phenomena under investigation. Qualitative research is individualistic in nature. Thus, in contrast to a quantitative and experimental methodology (Brocki & Wearden, 2006), the framework adopted provided a unique opportunity to rigorously examine parental accounts, delivering an analytical output that is informative, idiosyncratic and highly insightful in its ability to access the client's inner world (Smith, 2015).

In keeping with a qualitatively orientated approach, this study employed interpretative phenomenological analysis (IPA) (Smith, Flowers & Larkin, 2009). IPA was originally developed in the field of psychology as an experiential qualitative methodology. The first IPA research emerged in the mid 1990s (Smith, 1996), with the intention of studying experiential phenomena, and the meanings individuals assign to events in their life (Brocki

& Wearden, 2006). From 1996-2008, the total number of published studies reporting the use of IPA was 293 (Smith, 2011), although the IPA corpus is growing steadily, particularly in the areas of health-psychology, and illness research (Smith, 2015).

IPA theory

As a framework, IPA draws heavily upon the theoretical principles of phenomenology, hermeneutics, and idiography (Smith & Pietkiewicz, 2014). Phenomenology, a core idea of the philosopher Edmund Husserl (1859-1938) is interested in the study of experience, from an individual's perspective. The aim of phenomenology is to understand how events are perceived and talked about from a personalised perspective. A phemonenological approach follows a critique of positivist science, as it proposes that deductive, nomothetic methods of inquiry are insufficient to understand the lived reality, and complexity of individual experience (Larkin, Watts, & Clifton, 2006). Rather, the researcher works towards, as much as possible, understanding the meanings or "essence" of what is being said, from the perspective of the person being studied (Smith et al., 2009).

The views of Husserl were also developed upon via Martin Heiddiger (1889-1896), a German philosopher and freethinker who was primarily concerned with the study of 'hermeneutics'. Hermeneutics is an existentialist theory of interpretation (Larkin & Thompson, 2012). In the hermeneutic philosophical tradition, pre-suppositions are considered a key part of human existence (Dowling, 2012). Man develops an understanding of the world based on his own subjective leanings, which are influenced by the particularities of his own historical, cultural and linguistic environment. The influence of Heiddegerian hermeneutic phenomenology on IPA is made clear through the emphasis placed on the role of the researcher as an active 'sense making organism', who makes his own interpretation of the participant's interpretation of their experience. Smith & Osborn (2007) refer to this experiential process as the 'double hermeneutic'; one is ultimately engaged in a two-way effort to make sense of the participant, who is at the same time trying to make sense of their world.

Finally, idiography, refers to ‘an in-depth analysis of individual cases…in their unique context’ (Smith & Pietkiewicz, 2014, p.8). A detailed understanding & interpretation of each case is needed, prior to the development of broader, more general themes. The aim in IPA is to learn about the experiences of individuals, before interpreting shared meanings and patterns across cases (Brocki & Wearden, 2006).

Choice of IPA

The influence of phenomenology, hermeneutics and idiography on IPA is made clear through IPA’s idiographic commitment to the study of experience, and an explication of individual cases, within an interpretative framework (Smith et al., 2009). In this study, IPA was selected as the methodology of choice. Contrary to other competing methodologies, IPA focused more closely on studying experiences at the individual level.

Due to the sensitivities of this project it felt important to learn how parents think and feel about storytelling from a personal perspective. Hence, rather than conducting a thematic analysis or a grounded theory, which involves searching for essential structures (Braun & Clarke, 2013) or a unifying theory (Strauss & Corbin, 1997), IPA allowed for a closer analysis of the experiential data, both within and across each case. The use of the IPA framework, through its personalised emphasis, allowed greater space for idiographic variation than pattern based discourse analysis, which traditionally seeks to interpret the way that broader power relations and other socially available linguistic resources influence the production of knowledge (Fairclough, 2013). Indeed, such approaches are also common to narrative analysis and linguistic anthropology, which typically aim to examine the role of discourse and identity within everyday life. Unlike these approaches, the IPA method is individually orientated, and thus more able to account for the emotionality of topics that are deemed complex and deeply personal (Smith, Stephenson, & Quarrell, 2002). It allows for a sensitive exploration of affectively laden issues from a phenomenological perspective and, as such, IPA was considered appropriate for a study involving parents of children with LLCs and LTCs.

IPA publications in health psychology are also numerous (Smith, 2011). Hence, the health psychology emphasis of the project aligned naturally with the use of this methodological approach. Indeed, IPA's idiographic, experiential stance is crucial for understanding under-researched areas in illness. In 2011 it was also recognised that 24%, (N=64) of the published IPA corpus examined illness-related topics (Smith, 2011); thus signaling a congruity between the use of this method, and subject of the empirical paper (Chapter 3).

Epistemology

IPA is explicitly concerned with the construction of meaning, and how it is informed by an individual's personal and social world (Frost et al., 2010). IPA has a particular regard for the *person in context* (Larkin et al., 2006). The empirical paper was therefore undertaken from the contextualist epistemological standpoint (Madill, Jordan, & Shirley, 2000), and had a particular interest in the way that knowledge is local, provisional, and guided by the social and relational milieu (Madill et al., 2000). The understanding that reality is both subjective, and socially negotiated was reflected through the interpretative nature of the analysis and the adoption of a relativist ontology, as this is IPA consistent, and suggests a view of the world that is highly nuanced and guided by the diverse ways individuals understand and interpret events in their lives (Huws & Jones, 2010). The researcher interpreted the participant's interpretation of their experience (Smith, 2015), discussing potential differences in the phenomenon that arose when storytelling was undertaken in or outside the hospice context. Furthermore, as the contextualist position implies a non-foundational view of knowledge, in which reality is contextually based (Braun & Clarke, 2013), the researcher (T.M.) remained aware of his theoretical preconceptions and sought to maintain a more open-minded stance during the research through the use of a reflective diary. The first author's experiences, working in a paediatric service, visiting the hospice sites and reading widely around the topic of PPC, were also able to bring him closer to the phenomena under investigation and aid recognition of the way that social and cultural influences would inevitably impact on the results obtained.

Recruitment

Small, homogenous samples are favoured in IPA research (Smith et al., 2009). This study sought to recruit a relatively uniform demographic, (parents, from a single hospice charity), as it was thought that this homogeneity would allow for a more meaningful understanding of experiences within a particular context (Spiers, Buszewicz, Chew-Graham, & Riley, 2018). In the empirical paper, a limited number of parents (N=8) were purposively recruited from a small-scale charitable hospice organisation. The recruitment strategy is outlined in Chapter 3.

Sample

A total of 8 participants were recruited. All (n=8) were a biological parent of a child with LLC/LTC. Parents in this study were of working age (25-54), and were predominantly University educated, either to undergraduate (n=5), or postgraduate (n=1) level. One parent had GCSE/O-Level qualifications (n=1) and another did not have any educational qualifications (n=1). All the participants were White British, native English speakers (n=7), with exception of one Asian parent (n=1), who spoke English fluently as a second language. The children of these parents with LLC or LTC (n=7) had a mean age of 5.8 years.

The recruitment of 8 participants is appropriate (Robinson, 2014). Academic authors in IPA research typically recommend a small sample size, involving up to eight participants (Smith & Pietkiewicz, 2014). Particularly in clinical psychology doctorate theses, a small sample size of 6-8 is recommended in order to maximise feasibility (Turpin et al., 1997). The recruitment of this sample allowed the principal investigator to identify convergent and divergent themes across the sample (Smith, 2011), while ensuring that each individual's idiosyncratic experience was thoroughly reported within the research.

Interview

Semi-structured face-to-face interviews (n=8) were conducted privately, 1:1, in a quiet location of the participants preferred choice. Participants typically chose to be interviewed at their home address (n=7), although one participant was interviewed at his workplace for the reason of personal convenience (n=1). Interviews ranged from 45-79 minutes in length (mean=58 minutes) and were audio-recorded using an Olympus DS-2500 dictaphone.

Each interview was completed in accordance with the principles of IPA (Smith et al., 2009). Upon meeting, the primary researcher (TM) provided an explanation of the study, the voluntary nature of participation, and the right to withdraw from the study at any time, without giving any reason, until ≤2 weeks post-interview. Participants were given the opportunity to ask questions about the study and self-select a number of pseudonyms for themselves/their family for use in the study write up. Data storage, according to Data Protection Act (1998) principles was made clear.

All parents understood the project and the interview procedure before written informed consent was obtained. Participants completed a short demographic questionnaire, and were subsequently asked a series of open, expansive questions from an interview topic guide (appendix P). Six questions were contained within the topic guide, as recommended by Smith et al (2009), and all questions were checked by hospice staff/service-users and university staff prior to use. Questions moved gradually from having an initial descriptive focus, to a more analytical tone over time, as the there was a need to help the participant ease into the interview (Smith et al., 2009), and feel comfortable discussing the more emotive aspects of their experiences with a stranger (Dempsey, Dowling, Larkin, & Murphy, 2016). In this study the parents interviewed were considered experiential experts. Therefore the researcher assumed the role of an *active listener* (Smith et al., 2009), who followed the lead of the participant and deviated from the topic guide when needed to explore intriguing lines of inquiry as they emerged. Participants were provided with space to answer each question in detail. Therapeutic pauses gave rise to richer, fuller responses within the dialogue, while also cultivating rapport across the each of the interviews conducted. As the interviews progressed the researcher continued to note his emerging ideas in the reflective diary and remained aware of the need to resist engaging in preliminary data interpretation (Smith et al. 2009), thus allowing him to get closer to the essence of the individual's experience within the interview.

At points during the interview, the researcher summarised the conversation to ensure the participants' experiences were comprehended correctly (McCann, Lubman, & Clark, 2012). Breaks were offered to all participants, in order to ensure their comfort, safety and wellbeing. Once the interview concluded, parents were given the option to receive a further information sheet with telephone numbers for relevant services offering mental health support (Appendix Q). No adverse events were reported during interviewing; although detailed risk management/distress protocols (Dempsey et al., 2016) were constructed a-priori with a study collaborator at the hospice charity and were in place throughout recruitment (January-May) to fully support participant (Appendix R) and researcher wellbeing (Appendix R).

Analysis

In IPA, the analysis is a product of the researcher's interpretation of the interview data (Smith & Osborn, 2007). The interpretative activities of the researcher entail a process of sustained, analytic engagement with the interview material (Van Dijkhuizen, Clare, & Pearce, 2006), from which a series of themes, emphasising nuanced patterns of both convergence and divergence, begin to emerge (Smith et al., 2009). To ensure the interactive and immersive role of the researcher in the analysis process, all audio-recorded interviews were transcribed verbatim by the first author (T.M.) in Microsoft Word version

14.7, without use of an external transcription service. Audio files were replayed in Nvivo 11 (QSR International, 2013) at a reduced speed to facilitate transcription. Transcription was undertaken within 24 hours of each interview with the inclusion of regular breaks to optimise focus (Braun & Clarke, 2013).

Following transcription, the IPA framework of Smith et al (2009) was adhered to. The steps outlined by Smith et al (2009) are considered aid competency in the use of IPA. In this study the principle author was a novice qualitative researcher and therefore the use of a more practical, unidirectional IPA framework was considered preferable. The six flexible steps of Smith et al (2009) for conducting high-quality IPA were followed: (1) repeated re-reading of the transcript (2) initial commenting (3) theme development (4) identification of superordinate themes (5) moving to the next case (6) constructing superordinate themes via cross-case analysis.

Analytical approach

Transcripts were uploaded into NVivo 11 for analysis. Transcripts were initially analysed individually, case-by-case, as this is consistent with the idiographic emphasis of IPA (Smith et al., 2009). A cross-case analysis was only completed during the concluding stage of the analysis.

(1) *Repeated re-reading*: Initially, several readings of the first transcript were completed (Smith et al., 2009). Close engagement with the text involved paying careful attention to the significance and meaning of participant's words, in order to fully immerse oneself in the account (Mcormack & Katalinic, 2016). Efforts were made to consider the particular words or phrases the participant employed as this is thought to aid further intimacy with the data (Pringle, Drummond, & McLafferty, & Henry, 2011). Consideration was given to non-verbal aspects of the transcript (e.g. frequency of pauses) and notes from the reflective diary were reviewed in order to account for the possible implicit meanings within the text that could not be readily obtained from the participant's words alone.

(2) *Initial commenting*: Line-by-line comments were then added to the wide margin of the transcript (Smith et al., 2009). Points of verbal, descriptive, or conceptual interest were noted, along with any preliminary interpretation of the data. At this stage the aim was not to be overly definitive. No prescriptive rules were followed (Smith & Osborn, 2007; Smith et al., 2009), as the aim was to freely comment upon aspects of the text and emerging ideas as they came to mind. Appendix S provides examples of the comments added to six particular transcripts. The comments were added using the 'annotate' function in Nvivo11. The use of Nvivo11 proved advantageous in this regard, as the annotate function allowed for the comments to be viewed, reflected upon and further refined next to the relevant annotated text extract.

(3) *Theme development:* The researcher then returned to the beginning of the transcript (Smith & Osborn, 2007), and began to map connections between each of the exploratory comments (Smith et al., 2009). Various interpretative comments were linked together based on their conceptual similarity, before they were subsumed under a single label (i.e. theme). In keeping with the understanding of Smith et al (2009) themes constituted a simple phrase or word that broadly reflected the essence of the participants own words, as well as the interpretations of the researcher. At this stage, the researcher and his supervisor also discussed the possible emotional impact of undertaking the interviews, both as an ethical safeguard and to limit any issues of potential bias that might inadvertently impact upon theme development.

(4) *Identification of superordinate themes:* Once the themes were identified, connections between the themes were searched for (Smith et al., 2009). The themes were listed individually in NVivo11 and observed visually. Lines and arrows were further drawn within the reflective diary at this stage to make additional conceptual links between themes. Any themes that appeared to be interrelated were integrated within NVivo11 to form overarching superordinate themes. The superordinate themes were also checked against the original verbatim transcript, as this ensured that each of the superordinate themes had a clear link to the data (Smith et al., 2009).

(5) *Moving to the next case*: The aforementioned steps (1-4) were then repeated for all of the remaining transcripts. Each transcript was considered separately, in order to preserve the individuality of the account (Smith et al., 2009).

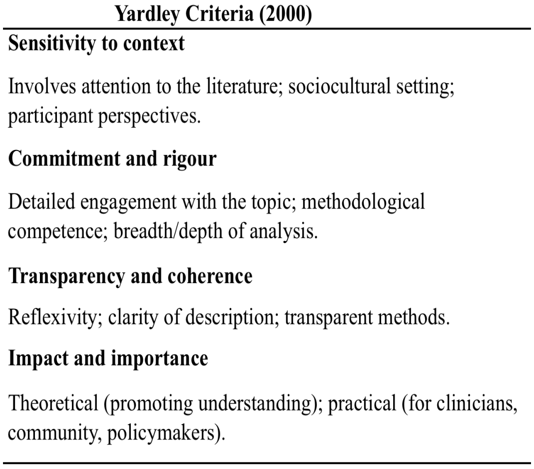
(6) *Constructing superordinate themes via cross-case analysis*: A comprehensive, consolidated list was then produced including all of the identified superordinate themes. Further integration occurred at this stage as superordinate themes from the transcripts were reviewed collectively (Smith, 2011); where conceptual similarities became apparent the superordinate themes were reorganised and clustered together (Smith et al., 2009). The superordinate themes were also checked by the primary supervisor at this stage and subjected to further reconfiguration (Smith et al., 2009), as is consistent with iterative process of IPA (Smith & Osborn, 2007). Following the initial write up, further reviews of the empirical paper were undertaken by two additional university academics experienced in IPA research through a Viva examination process. This led to further refinement of each superordinate theme and the further explication of implicit meanings within the data. Particular superordinate themes were relabelled subsequently and made clearer in terms of their link to storytelling within the write up (figure 1). The final superordinate themes were then reported in-text as part of the main body of the empirical paper (Chapter 3).

Initial superordinate themes (pre viva)	**Final revised superordinate themes (post viva)**
Sense of connection	Bonding with other parents through storytelling
Therapeutic compassion	Therapeutic storytelling to a hospice professional
Meaning-making	Storytelling as an educational tool
Fear of Judgment	Fear of others reactions to the story
Unhelpful repetition	Weariness through repetition of the story

Figure 1. Examples of superordinate theme relabelling from pre-post viva

Quality Criteria

The Yardley criteria are a number of flexible, open-ended principles that are thought to promote rigorous qualitative research. In this study, the four quality criteria of Yardley (2000) were applied, as this framework is considered congruent with IPA (Smith et al., 2009; Smith, 2011) and contains broad criteria that may be fulfilled flexibly, without rigid rules or prescriptions (Yardley, 2000). The Yardley criteria, outlined in figure 2, are a) sensitivity to context b) commitment and rigour c) transparency and coherence d) impact and importance.



Yardley Criteria (2000)

Sensitivity to context

Involves attention to the literature; sociocultural setting; participant perspectives.

Commitment and rigour

Detailed engagement with the topic; methodological competence; breadth/depth of analysis.

Transparency and coherence

Reflexivity; clarity of description; transparent methods.

Impact and importance

Theoretical (promoting understanding); practical (for clinicians, community, policymakers).

Figure 2. Yardley Criteria (2000)

Sensitivity to context

Yardley (2000) argues that an in-depth knowledge of the participants socio-cultural context is needed to promote high quality research. In order to maintain sensitivity to the particularities of parents' experiences (Smith et al., 2009), the researcher obtained feedback from parents regarding the recruitment materials/interview procedure, and combined this with a simultaneous practical effort to immerse himself in the participants 'local world' (Holloway & Galvin, 2016), via personal engagement in the sociocultural milieu of the hospice (Larkin, 2018). This process involved attending the hospice sites and conversing with parents directly. Following an invitation from staff, (a) a parent & toddler group, (b) a fathers group and (c) a bereavement group were attended. Speaking to parents in these groups helped to contextualise the interview data, thus allowing a more detailed, ethnographically sensitive analysis to be conducted (Hansen & Trank, 2016). Theoretical sensitivity, was also considered an important part of this process (Yardley, 2000), and was demonstrated by the researchers efforts to ground oneself intellectually in the philosophy of IPA (Smith et al., 2009), before conducting a detailed analysis according to its core principles.

Commitment and rigour

The criteria of commitment and rigour refer to thoroughness in the data collection-analysis process (Yardley, 2000). In this study, commitment and rigour were enhanced through “prolonged engagement” with the informants (Shenton, 2004). Via lengthy participant interviews (Mean duration =58mins) and a detailed, case-by-case approach to the study of each transcript, the researcher was able transcend superficial interpretations of the data (Yardley, 2000). Nuanced, multilayered understandings of the participants’ life world emerged, as the researcher behaved interpretatively, reviewing the transcript multiple times in an effort to try and uncover implicit meanings in the data. The researcher worked alongside a more senior academic supervisor in this process to check the themes as they emerged, thus ensuring they were credible and informed by multiple perspectives (Pringle et al., 2011). Repeated consultation with the supervisor ensured that all the superordinate themes were decided upon using a consensus approach, thus further enhancing the rigour of this empirical study.

Rigour is also demonstrated through the development of competency in the methods used (Yardley, 2000). As the researcher was initially new to IPA, extracurricular attendance at various groups facilitated the development of the competencies needed to conduct a phenomenological analysis. The researcher attended NVivo training, as well as a regional online IPA forum (http://www. ipa.bbk.ac.uk/discussion-group) and a UEA ClinPsyD Qualitative Discussion Group to gain additional insights into topic and build the repertoire of skills required for the completion of a high quality IPA project.

Transparency & coherence

Transparency and coherence refers to the rhetorical persuasiveness of the argument presented (Yardley, 2000). The argument described should have plausibility. In order to ensure the researcher worked from accurate data, a ‘member validation’ procedure was completed, in which consenting participants (n=4) were sent a copy of their verbatim transcript and asked to confirm its accuracy. Member validation is intended to verify information. It ensures the preliminary data is “accurate” according to the understandings of both the participant and researcher (Bygstad & Munkvold, 2007). The intention, in this case, is to ensure the participant’s experiences are represented in credible terms (Thomas, 2017). All participants consenting to this process (n=4) confirmed their transcripts were accurate, thus reflecting the way that parents were given influence, and actively engaged in the analysis process after interview. Once ‘member validation’ had been completed, reciprocity between the researcher and participants continued, as the interpretations of participant’s experiences were reported in-detail, using rich, thick verbatim quotes. In-depth descriptions of the storytelling experience via the participant allowed the reader to verify the legitimacy and coherence of the main analysis (Cresswell & Miller, 2000), thereby further enhancing trustworthiness.

A reflexive footing in qualitative research involves appreciation of the issue of *positionality* (Walker, 2013). This refers to a consideration of the way that ones personal assumptions or beliefs could potentially influence all aspects of the recruitment, data collection and analysis processes. Maintaining an open, reflexive stance is considered crucial in qualitative research, thus epistemological reflexivity was embedded into the study, through the use of a researcher led reflective diary.

The reflective diary constituted a tool for effectively considering the impact of the researchers position and interests on the study. In order to ensure a continuous process of hermeneutic reflection, the diary was kept on an ongoing basis (2016-2019; Jootun, McGhee, & Marland, 2009). Early preconceived ideas, or emerging personal thoughts relating to the interviews were included in the diary, in order to “bracket” the immediacy of ones assumptions, and therefore remain more open to the participant data in its original form (Tufford & Newman, 2012), An exemplar extract from the reflective diary, following the first interview, is outlined:

“The parent had positive beliefs about the hospice so perhaps was more

willing to participate. I wasn't sure during the interview how she felt about talking to me…my first impressions were that she was intelligent and articulate, which perhaps meant she was better able than some other individuals to tell her story. The parent was concerned about storytelling in public but seemed less concerned about telling the story in private".

"I was also aware that she seemed quite tired during the interview. I wondered if this impacted her ability to answer some of the questions. Towards the end of the interview I found myself thinking about illness in my own family, and I wondered if this had influenced my decision to take on this project".

Extracts from the diary were reflected upon subsequently in research supervision to maintain this open stance. Remaining aware of the impact of these beliefs and discussing them with an experienced supervisor, helped the researcher to separate his personal views from the research phenomena (Jootun et al., 2009), and thus limit their influence on the study findings.

Impact & importance

Finally, Yardley (2000) suggests that research should be judged according to its impact and utility. Research should have clear implications that extend beyond the study itself to impact upon the beliefs or actions of relevant stakeholders (Yardley, 20000). Results for the study were therefore theoretically novel. They were also designed to inform PPC service practice and systemic provision in hospice services.

In order to facilitate meaningful change through this study, the results will be circulated at meeting of key stakeholders at the children's hospice charity in 2019. Efforts will also be made to publish the findings in the journal of 'Psychology & Health', and present the outcomes at the forthcoming UEA Research Conference (2019), as it is important to ensure the wider potential impacts of this research are realised (Brownson, 2017).

5.

Extended Discussion

The extended discussion reviews the findings presented in this thesis. It discusses the systematic review results (Chapter 1), and seeks to consider links between the theoretical ideas presented in the bridging chapter (Chapter 2) and the empirical research paper (ERP; Chapter 3). Strengths and limitations in the thesis are discussed. Finally, clinical and research implications are explored.

Systematic review & ERP

A systematic review was conducted. It provided evidence that parents experience both positive psychological change and growth, alongside distress when caring for a child with LLC or LTC (Chapter 1). One therapeutic experience that is believed to have diverse, positive implications for emotional health and wellbeing in this population is storytelling (Chapter 2; Pennebaker, 2000). The ERP (Chapter 3) therefore investigated the experience of storytelling in parents of children with LLC and LTC.

Positive and negative experiences of storytelling were reported in the ERP. Parents predominantly felt that storytelling had a psychologically helpful quality and value within the context of a children's hospice, especially when stories were shared with other hospice parents and professionals ('bonding with other parents through storytelling'; 'therapeutic storytelling to a hospice professional'). Positive encounters also arose through parental efforts to educate the wider community ('storytelling as an educational tool'). However, parents predominantly reported negative experiences outside the hospice, for example when telling their story in everyday encounters or with other healthcare staff. Here, they were fearful of judgment ('fear of others reactions to the story') and could feel dissatisfied in their repetitive interactions with statutory HCPs ('weariness through repetition of the story').

These findings highlight the importance of social context in guiding storytelling as an experience. Whilst the hospice may function as an empowering, supportive context for storytelling, from which positive experiences typically emerge (Pennebaker, 2000), a socially constrained environment[7] is more likely to impede psychological adjustment and contribute to negative experience (Kolokotroni, Anagnostopoulos, & Hantzi, 2018). By drawing upon the theoretical ideas presented in the bridging chapter, the next section discusses the relational dynamic between social context and storytelling in light of the ERP findings.

Social context & storytelling

Storytelling experiences in the present study were informed by the social and relational milieu (Marino, Child & Campbell Krasinski, 2016). A larger proportion of positive experiences that were perceived as psychologically helpful and therapeutic were reported within supportive and empowering contexts (Pennebaker, 1997). Whilst, in the absence these conditions, positive encounters were not clearly evidenced, and some evidence of negative experience via storytelling was observed.

Mothers and fathers (n=8) of children with a wide range of LLCs and LTCs participated and spoke of a cathartic dimension to the storytelling experience. Where supportive social conditions were evident during storytelling (‘bonding with other parents through storytelling’; ‘therapeutic storytelling storytelling to a hospice professional’), or the parent assumed a position of social dominance by educating others (‘storytelling as an educational tool’) this seemed to result in evidence of interpersonal closeness, and other transformative feelings of wellbeing. This points to the importance of social and relational context in informing the storytelling experience (Lepore, 2001). Within the supportive and empowering context of the hospice, parents appeared to encounter feelings

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[7] Social constraint is defined as “any social condition that causes an individual to feel unsupported, misunderstood, or alienated when disclosing their concerns” (Cordova et al., 2001, p.706).

of closeness to their peers and experienced trusting therapeutic relationships with staff. Through educating others, they also noted diverse life changes, including feelings of greater pride, hope and self-efficacy. The extent to which is storytelling is experienced as having positive implications (Pennebaker, 2000) may therefore be influenced by ones perception of the social context, and whether it is considered supportive or sanctioning of storytelling as a social action (Mannell, Ahmad, & Ahmad, 2018). In keeping with the theoretical literature concerning self-disclosure (e.g. Lepore, Silver, Wortman, & Wayment, 1996), receptive social networks seemed to aid cognitive-emotional processing and facilitate storytelling (Koutrouli et al., 2016; Pennebaker, 2000). Meanwhile, the freedom to disclose one's story in an empathic environment allowed for further potential benefit finding, meaning making and positive reappraisal (Cordova et al., 2001).

Storytelling experiences were socially informed. Parents of children with LLC and LTC benefited from telling their story to a responsive, empathically engaged audience. The early identified themes (bonding with other parents through storytelling'; ''therapeutic storytelling storytelling to a hospice professional') are consistent with the wider literature on narrative theory (Pennebaker, 2000) and psychotherapy (Hardy & Sumner, 2018) as they demonstrate that speaking in a safe environment with likeminded individuals may help to facilitate bonding and disclosure. Stories perhaps became a means through which individuals were able to integrate new knowledge into their life narrative (Harber & Pennebaker, 1992) and develop a more normalised understanding of their experiences within a group context (Senehi, 2015). They were also able to build closer relationships with their peers via the hospice charity and experience a sense of acknowledgment from a number of professionals (e.g. counselors) within the context of a therapeutic dialogue (Caddick et al., 2015). Storytelling through the hospice often represented a psychologically therapeutic and positive experience, especially as it provided broader opportunities for validation and social support. The provision of validation and social support is widely noted as being an important ingredient in the experience of storytelling (Pennebaker & O'Heeron, 1984), which may help to facilitate healing (Wampold, 2018) and emotional catharsis (Gladding

& Wallis, 2010).

Parents also clearly made 'meaning' through narrative, as they felt able to shape public discourse through the disclosure of their story ('storytelling as an educational tool'). Within the hospice context and the community a number of parents became more aware of their oratorical skills (Heath, Farre & Shaw, 2017) and used their story to educate the layperson (Rafferty & Sullivan, 2017). Storytelling from the empowered social position of being an "educator" provided a context for meaning making (Graybeal. Sexton, & Pennebaker, 2002). Parents derived feelings of wellbeing (e.g. pride) and a sense of purpose from the opportunity to inform the wider public. This is in keeping with the theoretical understanding that storytelling can help an individual to make sense of, and find benefit in their experience (Bruner, 1991; Pennebaker, 2000; Stapleton & Wilson, 2017). Parents challenged the attitudes of others through the storytelling experience, and used the story for fundraising, which in line with related research, contributed to a sense of agency and influence (Cadell et al., 2012).

Nevertheless, negative experiences were also reported in the ERP, particularly where social constraints had the potential to impede storytelling. In themes four and five ('fear of others reactions to the story'; 'weariness through repetition of the story') "storytelling" outside the hospice could also prove problematic. Parents worried about the reactions of friends, family members, and strangers to the story, which detracted from their ability to speak openly. Evidence of this anxiety is contrary to the theoretical understanding that storytelling is a "cathartic" act (Pennebaker, 2000), or an avenue through which feelings of transformative experiential benefit typically emerge (Hemenover, 2003). In the absence of perceived social support, and the possibility of potential judgment, storytelling also seemed to be equated with a feeling of fear. This accords with narrative literature indicating that anxiety may persist (Wise, Marchand, & Roberts, 2018) or prove greater than any sense of positive experience in the context of storytelling (Lancaster, Klein, & Heifner, 2015). Pennebaker and colleagues argue that linguistic expression is unequivocally therapeutic (Hemenover, 2003; Pennebaker, 1997, 2000).

Therefore, their argument primarily emphasises storytelling as an experience that has positive implications for trauma management (Pennebaker, Colder, & Sharp, 1990). Investigations examining the adverse implications of storytelling are rare. However it has been noted that that negative experiences may arise, and even outweigh the positive impact in particular cases, especially where conversational opportunities for storytelling are poorly responded to (Yan & Bresnahan, 2018), or limited in scope (Prevatt & Desmarais, 2018).

The findings of this study suggest that storytelling is not solely associated with experiences that are cathartic and positive, but also negative. Although studies rarely highlight a relationship between negative affect and storytelling (McQueen, Kreuter, Kalesan, & Alcaraz, 2011), dissatisfaction and fatigue may take precedence where perceived social support is low (Sylaska & Edwards, 2014). In the context of the present study this was perhaps particularly evident, as parents reported additional stressful and frustrating examples of telling their story repeatedly to HCPs (‘weariness through repetition of the story’). Parents felt their interactions with statutory healthcare practitioners were often repetitive, which perhaps challenged their ability to make sense of, or encounter immediate positive benefit in their experience (Levetown, 2008). This is also consistent with the understanding that the quality of the relational dynamic in PPC settings, and perceived reactions to disclosure (Martin et al., 2018), influence the extent to which positive experiences are likely to be reported (Kuttner, 2007). Empathic listening/support is thought to create a sense of intimacy in storytelling (Shea, 2018), providing routes into catharsis and positive reframing of the trauma (Hibbin, 2016). Alternatively, if audience engagement with the story is poor, and individuals perceive others as disinterested, this inhibits cognitive-emotional processing and the increases the likelihood of dysphoria (Lepore, 2001). Experiences in the present study were similar. They were potentially guided by an interpersonal context, in which stress-related experiences and emotions were more closely associated with stories that were inhibited and sanctioned, rather than accepted and understood.

Strengths and limitations

Overall, the review and ERP results should be considered tentatively. This is due to recruitment from a single hospice charity and the inclusion of experiences obtained from only eight participants. The IPA study used a reflective diary to "bracket" the theoretical presuppositions of the researcher. While this limited the potential influence of theoretical biases and meant the researcher was perhaps better able to hold his personal assumptions in abeyance (Bendassolli, 2014), it possible that the prior knowledge derived from the systematic review (Chapter 1) and theoretical apparatus (Chapter 2) made it more difficult to interpret the participants experiences authentically without drawing upon this *a-priori* content during the analysis. The topic guide and related analytical outcomes did not map linearly onto the earlier described theory however (Pennebaker, 1997, 2000), which would appear to provide evidence of effective bracketing.

Participants in the ERP and review study were keen to share their experiences. This may have introduced selection bias (Robinson, 2014). Participants frequently self-disclosed positive experiences relating to the storytelling via the hospice. In turn, this appears to indicate that parents' with subjectively negative experiences were liable to be underrepresented. The transferable parameters of the findings could have been enhanced through the inclusion of further diverse perspectives, especially involving recently bereaved parents, and those who that had encountered suboptimal "storytelling" experiences inside the hospice.

Recruiting male participants to qualitative studies is a recognised difficulty (Oliffe & Thorne, 2007). Fathers are significantly underrepresented in the PPC literature. Whilst their inclusion in this study (n=2) increases the overall transferability of the findings, a disproportionate number of the study participants were female (n=8). Few individuals of a non-white British ethnicity also participated (n=1). The involvement of only a small number of male, or ethnic minority participants in both the systematic review and ERP perhaps limits the wider theoretical and cultural applicability of the findings presented in the thesis. Such groups are especially difficult to recruit into health illness research (Aristazabel et al., 2015). Macdonald, Chilibeck, Affleck, and Cadell (2010) for example, found that approximately 18% of adults involved in PPC research were male, compared to 82% female. In the present ERP, a broadly similar demographic representation was apparent (20% male, 80% female). The systematic review provided a proportionally larger number of males (30%), although this was only achieved by virtue of the strict inclusion/exclusion criteria[8]. Traditional masculine ideology (Affleck, Glass & Mcdonald, 2012) and cultural socialization (George, Duran, & Norris, 2014) are hypothesised as being influential factors in dissuading individuals from participating in qualitative studies that involve emotionally laden dialogue (Kristensen & Ravn, 2015). Thus, it is possible that the thesis inadvertently limited inclusivity by virtue of its discursive focus upon storytelling in the context of child illness and health.

IPA data collection methods were adopted in the ERP (Smith, 2015). The use of 1:1 semi-structured interviews, along with a 6-item topic guide, enhanced credibility, through a consistent approach to interviewing (McCann, Lubman, & Clark, 2009). However, the decision not to collect further subsequent data at a later time point also limited a more fluid and time-sensitive analysis of personal lived experience. The static snapshot obtained via a cross-sectional interview methodology meant that it was not possible to determine the extent to which experiences fluctuated, or remained stable over time (Snelgrove, 2014). Fewer opportunities were provided to capture transformative understandings of this phenomenon. A qualitative longitudinal IPA approach was overlooked (e.g. Snelgrove, 2014), and in conducting the interview at a single time point, this limited opportunities to study nuanced individual and group perspectives in their temporal context (Jeffrey, 2018). Furthermore, in collecting data through single interviews, the researcher omitted further large-scale data validation strategies (e.g. IPA focus groups), and relied predominantly on his own analytical contribution and K.M. to explore the study data. The involvement of further experienced researchers in the research process could have improved

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[8] Studies had to include both male and female perspectives.

credibility, and allowed for additional richer data to support the claims being made (Shenton, 2004).

As a construct in the wider literature, "storytelling" is broadly defined (Palacios et al., 2015). The decision to study storytelling in broad terms limited dependability of the findings (Morse, 2015). Parents described storytelling in a range of settings, which perhaps also reflected a heterogeneous understanding of the phenomena under investigation. Given the homogeneity principle of IPA, the study could have been improved through a narrower analysis, focusing on a smaller number of rich, descriptive cases (e.g. 4-6), in a single context (Smith, Flowers, & Larkin, 2009) and could also have benefited from an improved literature base, from which a uniformly accepted definition of "storytelling" could be drawn. In depth multiple perspective approaches are becoming more common in IPA studies (Smith et al., 2009) and a multiple perspective IPA could have further enhanced the research by allowing for a focus on both the experience of both the storyteller and the listener in a unique context.

Despite these limitations, a strength of the study is the use of member validation. Interview respondents were given the opportunity to review their own transcript and comment upon its accuracy, thereby allowing for the remediation of any perceived errors in representing parents' experiences (Thomas, 2017). The use of this technique allowed for participant engagement in finalisation of the transcripts (n=4) and in turn, this increased the potential confirmability and credibility of the research process. As a participatory, collaborative and systematic approach to ensuring data collection, member validation is consistent with the principles of IPA (Smith, 1996). In IPA shared understandings are formed by actively including both the participant and researcher "voice" in the research process (Smith, 2015). Further member validation in the later stage of analysis, such as participant verification of the superordinate themes, is perhaps likely to have been of benefit and enhanced trustworthiness, as Smith (2007) suggests such a process of verification represents the extension of IPA's interpretative methodology (Smith, 2007; Pringle, Hendry, McLafferty, 2011).

Superordinate themes were considered by the second author (K.M), given her expertise in IPA. In the analysis, links between each meta-theme and the interview content were double checked via supervisory discussion with K.M. This helped to ensure the study findings were not guided independently by the personal biases of the first author (Shenton, 2004), but were considered tentatively, and were subject to further academic critique through recursive discussion involving an experienced secondary analyst. In order to maintain a link to the original data during this process, themes were cross-referenced against the original transcripts and were also described in extensive detail, with the inclusion of rich thick and verbatim quotes helping to provide further evidence of rigour.

A final strength is that the analyses, at the time of writing, were novel. The systematic review and ERP both focused on exploring the experiences of parents caring for children with severe illnesses (LLC/LTC) and included those with rare under-researched diagnoses. Themes were well represented across participants. Thus, the thesis research project met an ongoing need for inclusive research examining the experiences of parents of children with genetically rare progressive non-malignant LLCs and LTCs.

Research implications

Further studies relating to storytelling in the context of PPC are warranted. The storytelling experiences reported in the ERP were to the author’s knowledge, unique, and were drawn from parents attending a single children’s hospice charity. However, there remains a dearth of evidence in hospice settings (Dunbar, Carter, & Brown, 2018), upon which optimal care for parents can be based. A lack of evidence relating to paediatric hospice services has been linked to the ethical complexities of conducting research in PPC (Downing et al., 2015), a focus upon adult palliative care (Pentaris, Papadatou, Jones & Hosang, 2018) and limitations in research funding (Scott, Jindal-Snape, & Manwaring, 2018). Historically the total reporting rate (3%) for PPC studies in

UK palliative care research is low (Kumar, 2011). Few contemporary high quality studies (n=12) were also identified in the systematic review. The end result of this is that paediatric hospice care does not currently have a rigorous empirical knowledge base (Downing, Namisango, & Harding, 2018), from which principles of best practice for supporting parents can be derived. Only a small number of studies have explored psychosocial support for parents in a UK children's hospice context (Wray, Lindsay, Crozier, Adams, & Leeson, 2013), resulting in inconsistent access to therapy for these adults (Weaver et al., 2016).

As a consequence, it is incumbent upon researchers to promote an evidence base in this area to strengthen services (Downing et al., 2015), and develop the principles of holistic, family centered care, that can better address the needs of parents that are caring for children with complex illnesses. The present study suggests that storytelling offers psychologically positive and therapeutic experiences, especially when delivered in an institutionally supportive and empowering environment, which includes likeminded individuals or opportunities to influence others. Although storytelling in these particular contexts may prove cathartic for parents this evidence is the first of its kind. The majority of research in this field to date has only focused on the experiences of storytelling in paediatric cancer patients (Ghirotto et al., 2018). Thus, additional hospice centred storytelling research involving parents of children with LLC and LTC is likely to be of future value.

The present findings imply that further research is needed to explore the benefits of storytelling in a hospice. The first identified theme ('bonding with other parents through storytelling') suggested that storytelling provides parents' with feelings of closeness to their peers in the hospice. Parents noted that these experiences often arose in a group context. It may therefore be worthwhile undertaking a further hospice-based study that seeks to investigate the formation of peer relationships through storytelling, especially in groups, as little is currently known about what parents in PPC want from childrens hospice services in the UK (Wray et al., 2013) and which approaches to storytelling are deemed most preferable (NICE, 2016). In the present study storytelling in a group provided a means for receiving further practical and emotional support from ones peers. Thus there is a need for further research assessing the implications of peer related storytelling to help guide resource prioritisation and inform the systemic quality of care within the hospice.

In the second theme (‘therapeutic storytelling to a hospice professional’) parents spoke of the acceptance and understanding they experienced through storytelling to hospice professionals. However, there is a lack of research exploring staff-parent relationships in this context (Wilkinson, Croy & King, 2007). Although hospice staff and counselors were considered especially skilled in their ability to cultivate a therapeutic dialogue through storytelling, more research is needed to efficiently investigate the therapeutic strategies they implement for this purpose (e.g. active listening), and ensure that the professional qualities that are valued by parents during storytelling are fully understood. Further research in this area can expand awareness of good professional practice and help facilitate skilled responding to parents’ stories.

Parents in this study also employed storytelling for pedagogical purposes (storytelling as an educational tool’). Parents were seeking to be a catalyst for change as they derived meaning, purpose and emotional strength from telling their story to educate others. Although storytelling in an organised educational context constituted an empowering experience, the majority of healthcare studies reporting similar findings have focused on parental advocacy in children with autism (Burke et al., 2018) or learning disability (Hess, Molina, & Kozleski, 2006). Hence, they have not typically explored the role of parental storytelling in promoting an improved knowledge of paediatric palliative concerns (Rafferty & Sullivam, 2017). A further novel study could therefore be conducted that examines storytelling as an educational tool in parents of children with LLC and LTC through public speaking, fundraising, and media events in the micropolitical domain.

Contrary to studies focusing on the health benefits of narrative (Pennebaker, 2000), two of the themes in this study also indicated that parents face difficulties in their disclosure of the story, especially outside the hospice (‘fear of others reactions to the story'; 'weariness through repetition of the story'). The negative experiences of storytelling therefore require further investigation. An investigation of adverse experiences should be undertaken that considers the impact that fear of judgment and repetition as variables have upon the storytelling experience given their identification in two ERP themes (Ekberg, Bradford, Herbert, Danby, & Yates, 2015). Parents in the review literature also reported high levels of pre-existing psychological distress. Identifying potential harms in storytelling, as well as harm minimisation strategies in future will be important.

Additional storytelling research could also be undertaken with smaller, homogenous samples such as fathers, and persons from particular ethnic minority groups. The conduct of this thesis, and research in PPC more generally, is focused predominantly on a Western female focused sample (Colville & Gracey, 2006). This hinders our ability to offer culturally relevant care that takes into account the experiences of all members within the family system. As the systemic and cultural make up of families in the UK becomes increasingly diverse (Nazroo et al., 2018), it may be important to examine the storytelling experiences of other family members that are involved in supporting children with LLC and LTC. Studies, for example, have noted that foster carers (Wood, Simpson, Barnes, & Hain., 2010), grandparents (Heath et al., 2017), and siblings (Lane & Mason, 2014) all play an active role in providing paediatric care and support across the illness trajectory, although as yet, research exploring their experience from a "storytelling" perspective is lacking. This points to the need for a broader family orientated perspective and research emphasis in PPC storytelling, which will have progressive implications for the delivery holistic care.

Clinical Implications

The ERP suggested parent-to-parent support within a hospice setting is beneficial when administered via the medium of storytelling. As storytelling seems to function as a potential catalyst for positive relational experiences between parents in the hospice (‘bonding with other parents through storytelling’), it may be worthwhile offering parents access to a brief informal storytelling group, or access to a ‘peer coach’ role, where parents can use their story as a carer to share “trade secrets” and information with parents of newly diagnosed children (Tully et al., 2017). Attempts to match parents of children with similar illness conditions together on the basis of clinical similarity may also prove helpful. Parents in the ERP reported a preference for telling their story to parents facing similar diagnostic issues. Hence, storytelling could potentially be promoted among parents in a diagnosis specific context (Martin et al., 2018).

The study suggests that parents benefit from their story being heard by a compassionate supportive listener in a therapeutic context such as the hospice. In this setting counselors and staff were perhaps helpful in maintaining an empathic focus on the parents story. Professionals helped to cultivate a therapeutic dialogue that contributed to the alleviation of distress. Thus, one implication from this finding is that storytelling in a 1:1 context between parents and hospice staff could be further offered as a basis for promoting catharsis and psychological healing within the hospice. Indeed, studies have previously found that storytelling to a therapeutic professional can help individuals establish a richer, holistic strengths-based narrative (Guilfoyle, 2016), and allow clients to experience improved wellbeing in a collaborative non-pathologising professional context (AnjaBjorøy, Madigan & Nylund, 2016). Thus, additional storytelling engagement with a compassionate hospice professional may prove helpful for parents, by allowing the parent to receive acceptance, understanding and find emotional benefit in adversity (Lopes et al., 2014).

Parents also encountered other benefits from telling their story. In telling their story publicly, either via a hospice or in a community setting, parents were able to derive feelings of meaning and empowerment from their experience. To date few studies have noted the positive role that public education and advocacy may play in supporting parents of children with LLC and LTC (Caddell, Kennedy, & Hemsworth, 2012). However it may be that providing parents with a platform to educate others is helpful. According to consumer-based models of healthcare, activism and advocacy work has the potential to enhance public knowledge of paediatric palliative healthcare issues and enable parental empowerment (Rafferty & Sullivan, 2017). Furthermore it can also facilitate co-operation in addressing barriers to healthcare provision, such as navigating the PPC system effectively. While parents previously identified 'navigating the system' as a problematic in the review, educating individuals through storytelling in the ERP provided a way to make others aware of the types of challenges presented by paediatric illness conditions. In this respect findings highlight the importance of offering parents the opportunity to speak publicly in an educational capacity. This could be undertaken either inside a hospice or outside a hospice via teaching events. Parents in the ERP also encountered positive emotional experiences from educating others, suggesting further use of this practice is warranted.

It is important to note that parents were also concerned about telling their story to friends, family members and stangers, who were not perceived to have a well-informed understanding of paediatric illness conditions. Parents worried about telling their story outside the hospice. If parents activities are limited by anxiety, as well as other challenges identified in the review literature, (such as carer burden) this may prevent parents from optimizing the use of storytelling as an empowerment activity (Boshoff, Gibbs, Phillips, Wiles, & Porter, 2016). In order to manage barriers that may prevent parents from speaking more publicly, it may be important for professionals to work therapeutically with parents to manage their anxiety, or find flexible, time sensitive ways to promote storytelling, that are culturally and linguistically suitable for each parent. Given the the multifaceted difficulties of being a full time carer, service providers will need to demonstrate responsiveness to the challenges of storytelling in order to ensure parents voices are heard (Boshoff et al., 2016).

Professionals may also need to directly challenge negative public attitudes or misconceptions regarding PPC. Research has shown that there is a tendency in the UK to incorrectly equate hospice provision with end of life care (Price, McCloskey, & Brazil, 2018). In the wider literature laypersons report being "intimidated" by concepts of child illness and interpret referral to a hospice as representing a "covert message" of impending death (Pentaris et al., 2018, p.653). In the present study, parents also reported that their experiences of storytelling were attenuated by anxiety, as well as concerns regarding inappropriate questions and problematic interactions with individuals who did not always appear to have a detailed, accurate knowledge of PPC issues and their impact upon the wider family system. In order to address this problem, further public education may need to target particular lay demographics (family, friends, strangers), and resources may need to be devoted to exploring parents' previous difficulties disclosing their story in the wider community. If parents are inclined to censor or leave out important aspects of their story due to feelings of fear this may limit disclosure and the potential benefits of storytelling.

Furthermore, there is a need to address the way that parents in the study encountered dissatisfaction and fatigue as they repeated their story to statutory HCPs. In order to ensure that parents feel involved in a helpful HCP dialogue, retention of the story and empathic listening are considered important. In acute medical settings resource limitations and lack of staff continuity may lead to increased repetition of the story, as support for the parent is often fragmented (Ekberg et al., 2015). Subsequently, there is a need for communication training programs run by the General Medical Council (Christie & Glew, 2017) and the Royal College of Paediatrics and Child Health (2016) to emphasise retention of the story (Davies et al., 2003). Novel easily implementable ways for parents to share their stories with HCPs are also required. Thus it may be that services need to encourage the use of creative storytelling (e.g. through art, writing) to capture the interest of statutory professionals in the context of resource limitations and reduce verbal repetition of the story. Artistic approaches, such as 'beads of courage' and written aids which allow parents to "story" healthcare experiences using creative methods, were used by parents in the present study (see Chapter 3) and are likely to be of additional help in securing positive, engaging storytelling interactions with child health providers in the future.

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Appendices

Appendix A: Systematic Review Development

What are the experiences of parents when caring for a child with a life-limiting condition or life-threatening condition? A systematic review and narrative synthesis (PROSPERO registration no: CRD42017083265)

The proposal for the systematic review study above was initially made in June 2017 with the submission of the thesis proposal and received positive comment from the markers.

The review itself was written and completed by the end of December 2017 and sent to the primary supervisor. The paper by Bally et al (2018) was published in January/February 2018. Critical differences between this paper the systematic review exist.

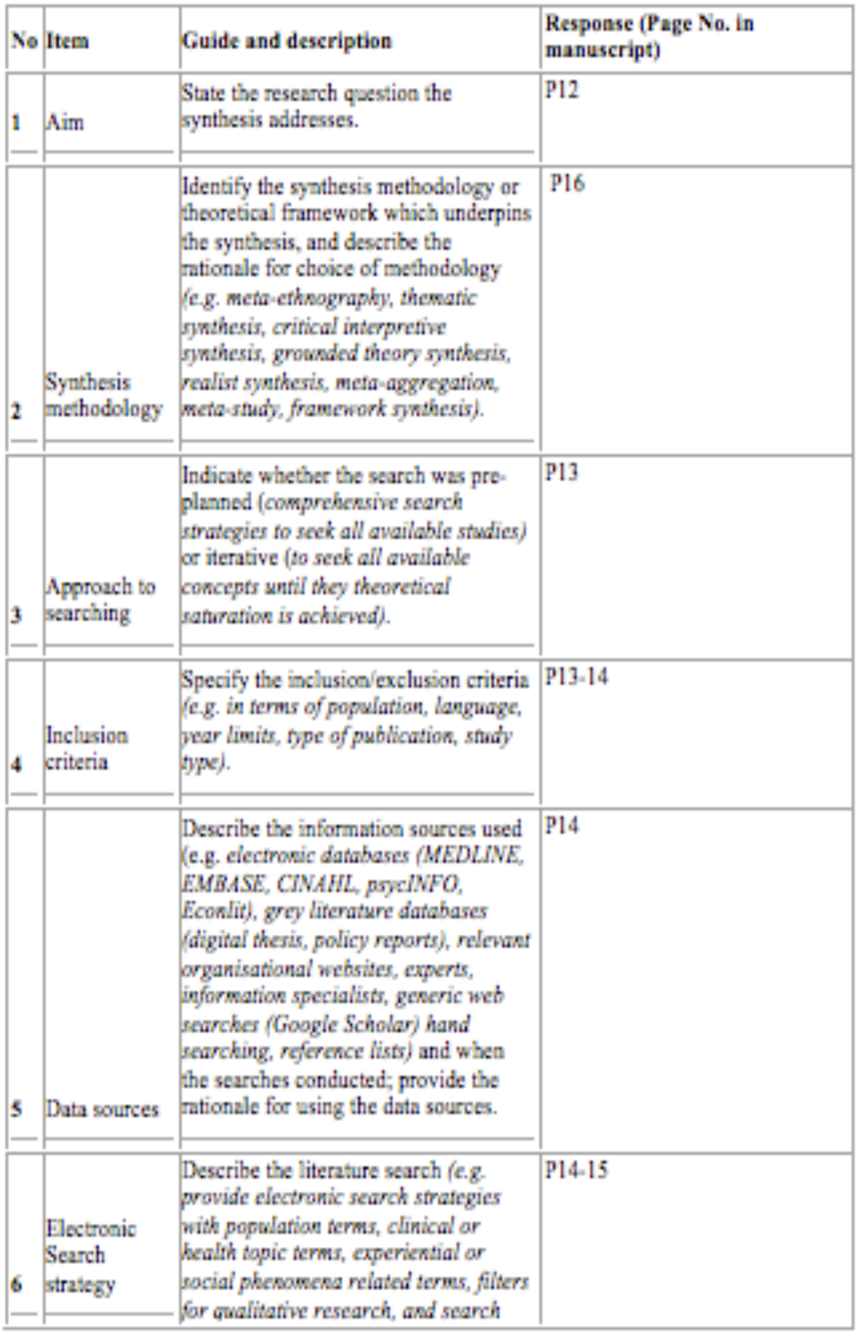
These include a) the fact Bally et al (2018) seek to explore the experiences of families, parents, grandparents, siblings, and children collectively. The findings from each cohort are integrated together thus making it difficult to differentiate the experiences of parents from those of the wider family. Unlike our study, the decision is made in their study to exclude studies in developing nations and there is c) no focus upon the primary construct of caregiving. It is d) unclear whether the Bally et al. (2018) study includes literature that is peer-reviewed, and e) meta-synthesis is employed to integrate the findings.

Bally et al (2018) also f) omit the majority of the recently published research in our review (e.g. Collins et al., 2016, Somanadhan & Larkin, 2016., Verberne et al., 2017, Hayles et al., 2015; Taleghani et al., 2012; Klassen et al., 2012) as well as a number of older studies (e.g. Wong & Chan, 2006, Davis et al., 2010, Flury et al., 2011; Patterson-Kelly et al., 2011). Unlike Bally et al (2018) g) our study omitted research deemed inadequately reported and h) focused on including studies that involved both mothers and fathers. This is recommended in the PPC literature as research in this field has repeatedly emphasised the importance of representing male/female experiences more equally (see Macdonald, Chilibeck, Affleck, and Cadell, 2010). Our review also reports a larger variety of themes, i) which are often different from those reported in Bally et al. 2018 and includes a more contemporary focus by including j) literature post 2014.

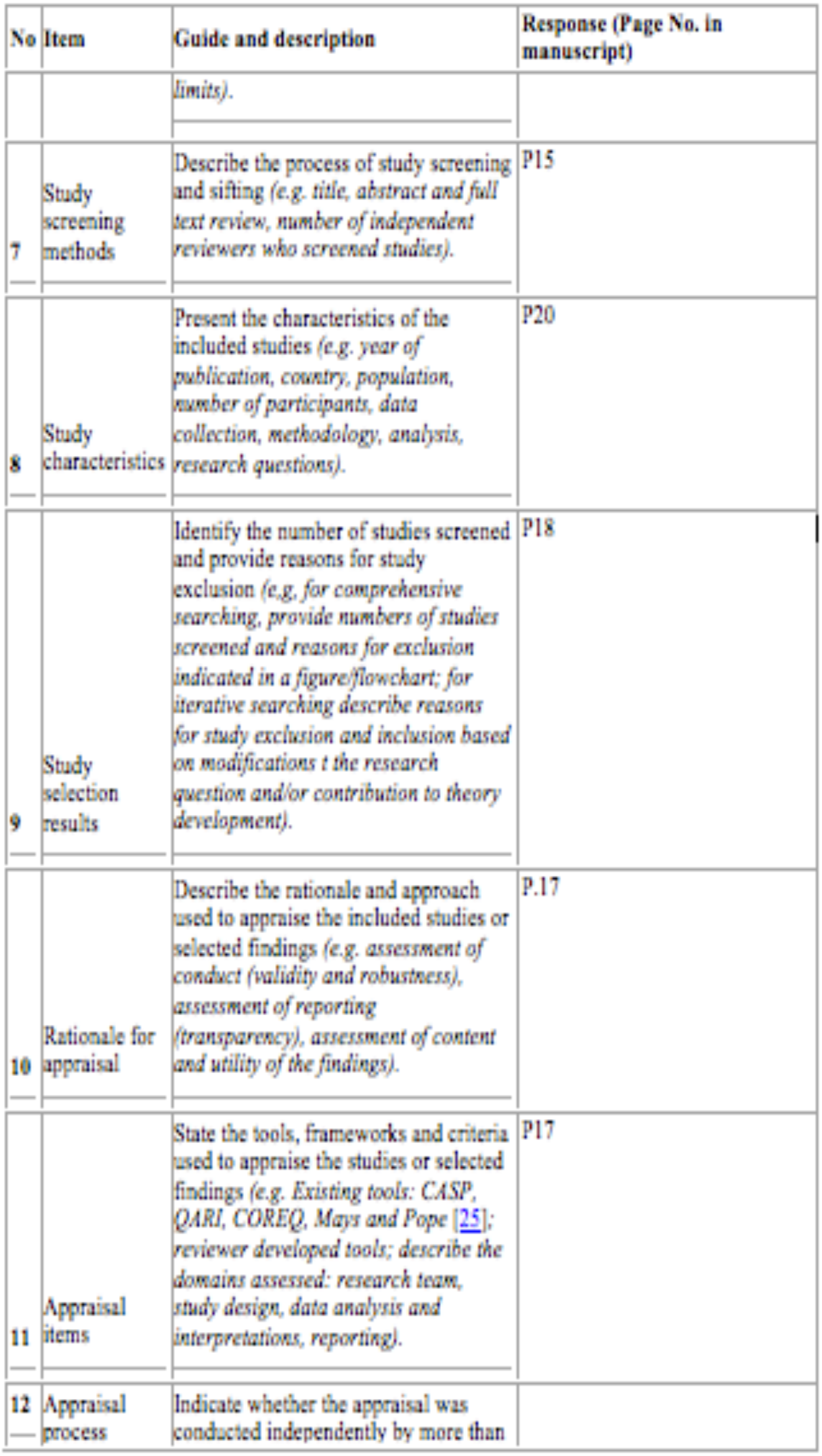
Personal correspondence with the journal editor of Health Psychology Review also confirmed that in light of the differences named above that the study above would be considered sufficiently novel to be considered appropriate for submission to the journal. Personal discussions with the second author Kiki Mastroyannopolou and another faculty member from the UEA ClinPysD programme also confirmed this.

Appendix B: Systematic Review Guidelines (ENTREQ, CRD, PRISMA)

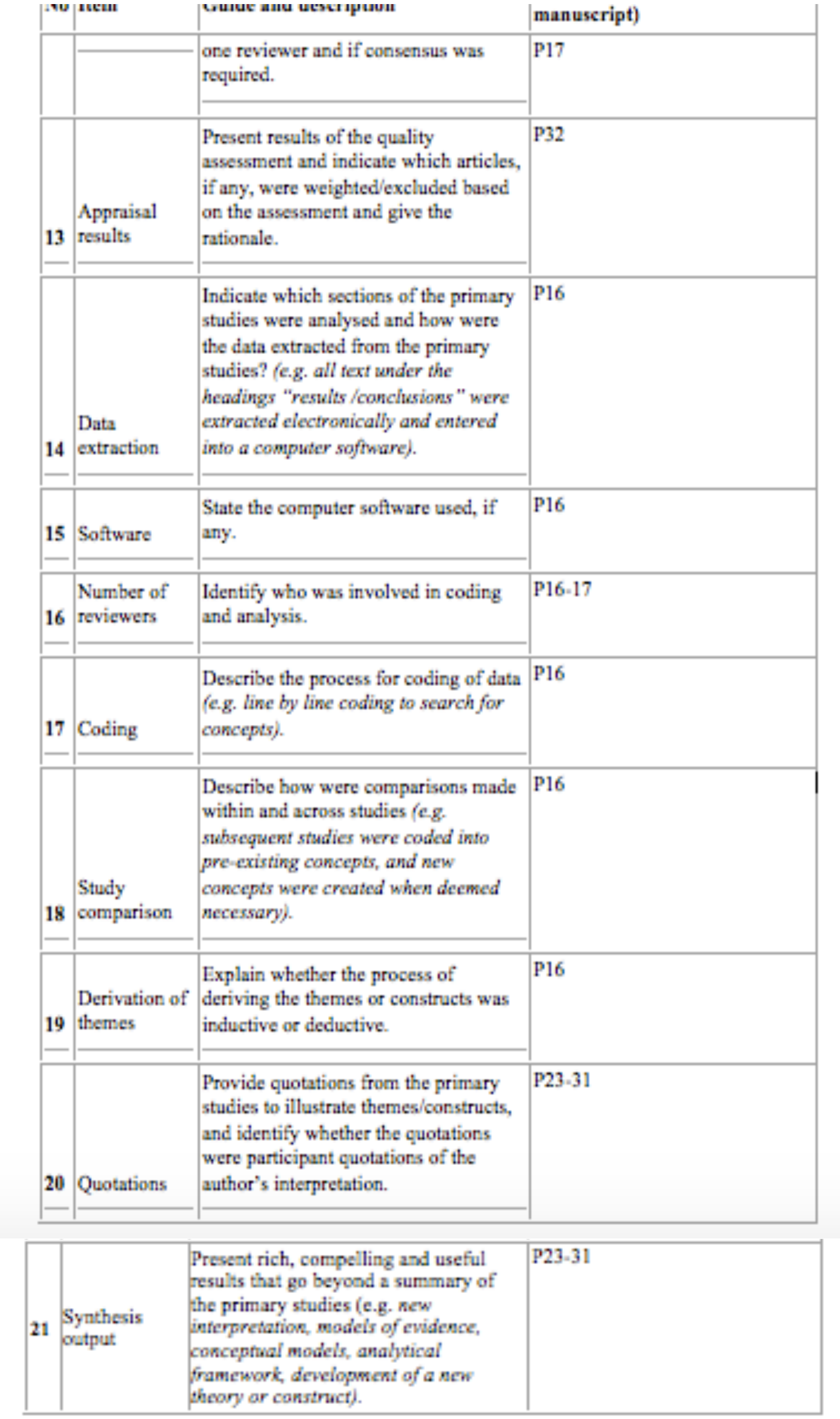
ENTREQ (Tong et al., 2012)



No	Item	Guide and description	Response (Page No. in manuscript)
1	Aim	State the research question the synthesis addresses.	P12
2	Synthesis methodology	Identify the synthesis methodology or theoretical framework which underpins the synthesis, and describe the rationale for choice of methodology *(e.g. meta-ethnography, thematic synthesis, critical interpretive synthesis, grounded theory synthesis, realist synthesis, meta-aggregation, meta-study, framework synthesis).*	P16
3	Approach to searching	Indicate whether the search was pre-planned (*comprehensive search strategies to seek all available studies)* or iterative (*to seek all available concepts until they theoretical saturation is achieved).*	P13
4	Inclusion criteria	Specify the inclusion/exclusion criteria *(e.g. in terms of population, language, year limits, type of publication, study type).*	P13-14
5	Data sources	Describe the information sources used (e.g. *electronic databases (MEDLINE, EMBASE, CINAHL, psycINFO, Econlit), grey literature databases (digital thesis, policy reports), relevant organisational websites, experts, information specialists, generic web searches (Google Scholar) hand searching, reference lists)* and when the searches conducted; provide the rationale for using the data sources.	P14
6	Electronic Search strategy	Describe the literature search *(e.g. provide electronic search strategies with population terms, clinical or health topic terms, experiential or social phenomena related terms, filters for qualitative research, and search*	P14-15



No	Item	Guide and description	Response (Page No. in manuscript)
		limits).	
7	Study screening methods	Describe the process of study screening and sifting *(e.g. title, abstract and full text review, number of independent reviewers who screened studies).*	P15
8	Study characteristics	Present the characteristics of the included studies *(e.g. year of publication, country, population, number of participants, data collection, methodology, analysis, research questions).*	P20
9	Study selection results	Identify the number of studies screened and provide reasons for study exclusion *(e.g, for comprehensive searching, provide numbers of studies screened and reasons for exclusion indicated in a figure/flowchart; for iterative searching describe reasons for study exclusion and inclusion based on modifications t the research question and/or contribution to theory development).*	P18
10	Rationale for appraisal	Describe the rationale and approach used to appraise the included studies or selected findings *(e.g. assessment of conduct (validity and robustness), assessment of reporting (transparency), assessment of content and utility of the findings).*	P.17
11	Appraisal items	State the tools, frameworks and criteria used to appraise the studies or selected findings *(e.g. Existing tools: CASP, QARI, COREQ, Mays and Pope [25]; reviewer developed tools; describe the domains assessed: research team, study design, data analysis and interpretations, reporting).*	P17
12	Appraisal process	Indicate whether the appraisal was conducted independently by more than	



No	Item	Guide and description	manuscript)
		one reviewer and if consensus was required.	P17
13	Appraisal results	Present results of the quality assessment and indicate which articles, if any, were weighted/excluded based on the assessment and give the rationale.	P32
14	Data extraction	Indicate which sections of the primary studies were analysed and how were the data extracted from the primary studies? *(e.g. all text under the headings "results /conclusions" were extracted electronically and entered into a computer software).*	P16
15	Software	State the computer software used, if any.	P16
16	Number of reviewers	Identify who was involved in coding and analysis.	P16-17
17	Coding	Describe the process for coding of data *(e.g. line by line coding to search for concepts).*	P16
18	Study comparison	Describe how were comparisons made within and across studies *(e.g. subsequent studies were coded into pre-existing concepts, and new concepts were created when deemed necessary).*	P16
19	Derivation of themes	Explain whether the process of deriving the themes or constructs was inductive or deductive.	P16
20	Quotations	Provide quotations from the primary studies to illustrate themes/constructs, and identify whether the quotations were participant quotations of the author's interpretation.	P23-31
21	Synthesis output	Present rich, compelling and useful results that go beyond a summary of the primary studies (e.g. *new interpretation, models of evidence, conceptual models, analytical framework, development of a new theory or construct).*	P23-31

CRD (2009)

Full guidelines available from:
https://www.york.ac.uk/media/crd/Systematic_Reviews.pdf

PRISMA (2009)

Section/topic	#	Checklist item	Reported on page #
TITLE			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	p.6
ABSTRACT			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	p.7
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known.	p.8-12
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	SPIDER criteria are used as an alternative to PICO, question is stated p12, and reference to the way that it is informed by SPIDER is on p.13
METHODS			
Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number.	p.13

Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	p.13
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	p.14
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	p.15
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	p.15
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	p.16
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	p.16
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	p.17- 18
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	NA - qualitative
Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I^2) for each meta-analysis.	p.16-17
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	p.17-18
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	NA
RESULTS			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	p.18
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	p.19-22
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item	p.32

		12).	
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	NA
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	NA
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	p.32
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	NA
DISCUSSION			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	p.33-36
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	p.35-36
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	p.37
FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	p.38

Appendix C: Health Psychology Review Guidelines

Full guidelines are retrievable from:
https://www.tandfonline.com/action/authorSubmission?journalCode=rhpr20&page=instructions.

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Your paper should be compiled in the following order: title page; abstract; keywords; main text introduction, materials and methods, results, discussion; acknowledgments; declaration of interest statement; references; appendices (as appropriate); table(s) with caption(s) (on individual pages); figures; figure captions (as a list).

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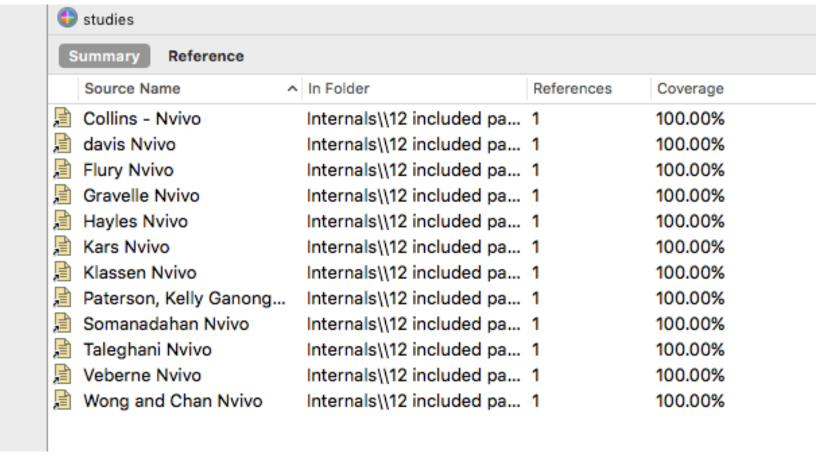
Appendix D

Stages of analysis and extracts from Nvivo11

(1) data familiarisation (2) coding (3) theme development (4) theme review (5) agreeing the final theme labels and (6) producing a final report.

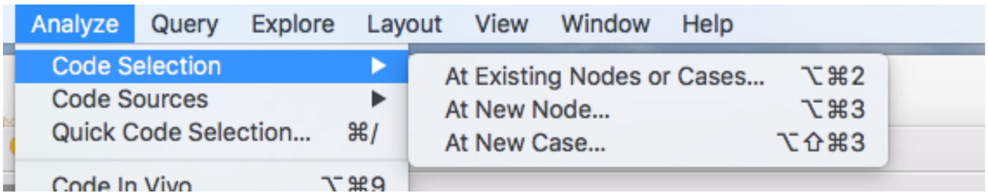
1). Data familiarisation: Firstly, the relevant data were extracted from each study and tabulated to create a preliminary synthesis (see table in main text).

2) Coding: To begin coding the results sections from the 12 studies were then imported into Nvivo. The extracted material from the results sections included all headings, participant quotations and secondary interpretations provided by the original study authors. The example image from Nvivo demonstrates that 12 sources (studies) were imported into Nvivo.

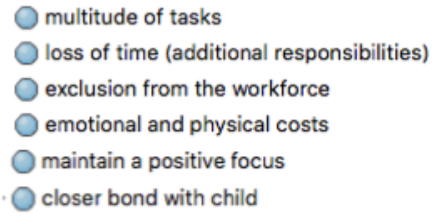


Source Name	In Folder	References	Coverage
Collins - Nvivo	Internals\\12 included pa...	1	100.00%
davis Nvivo	Internals\\12 included pa...	1	100.00%
Flury Nvivo	Internals\\12 included pa...	1	100.00%
Gravelle Nvivo	Internals\\12 included pa...	1	100.00%
Hayles Nvivo	Internals\\12 included pa...	1	100.00%
Kars Nvivo	Internals\\12 included pa...	1	100.00%
Klassen Nvivo	Internals\\12 included pa...	1	100.00%
Paterson, Kelly Ganong...	Internals\\12 included pa...	1	100.00%
Somanadahan Nvivo	Internals\\12 included pa...	1	100.00%
Taleghani Nvivo	Internals\\12 included pa...	1	100.00%
Veberne Nvivo	Internals\\12 included pa...	1	100.00%
Wong and Chan Nvivo	Internals\\12 included pa...	1	100.00%

Codes were then created in Nvivo11 to initiate the analysis. Codes were created by highlighting relevant sections of text in the results section of each study and using the code ‘at new node’ function in Nvivo 11.



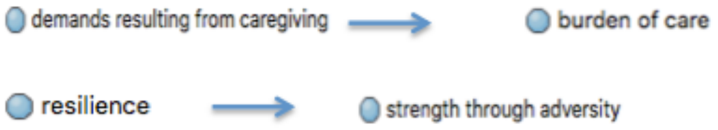
Codes consisted of a brief interpretative statement that reflected the underlying meaning or essence of a particular data extract. The coding process involved coding both the participants’ own quotations and the authors broader interpretations within the text. Examples of a subset of codes are provided below.



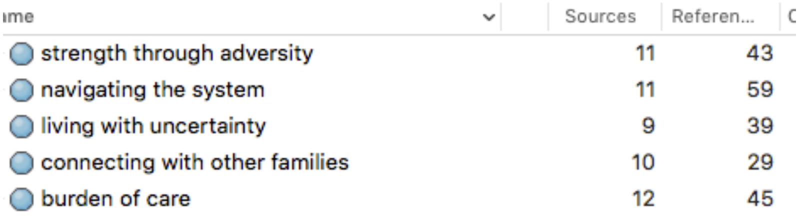
- multitude of tasks
- loss of time (additional responsibilities)
- exclusion from the workforce
- emotional and physical costs
- maintain a positive focus
- closer bond with child

3). Theme development: Clusters of codes were grouped together where appropriate to succinctly capture similarities across included studies. The clusters of codes were then relabelled to form candidate themes. For example, the initial codes mentioned (e.g. multitude of tasks, loss of time, exclusion from workforce, emotional/physical costs, positive focus, closer bond with child) were grouped together and relabelled to form individual candidate themes (demands resulting from caregiving, resilience). This process is outlined in the Nvivo image below.

The candidate themes were reviewed, reorganised and refined iteratively, until a coherent set of themes emerged that provided a clear and comprehensive representation of the data corpus. Through this process the candidate theme ‘demands from caregiving’ transitioned to become the theme ‘burden of care’ and the candidate theme ‘resilience’ became ‘strength through adversity’.



4. Theme Review: Each theme was assessed for homogeneity, frequency of data and relevance. Throughout this process the themes were checked repeatedly. Themes were subjected to repeated discussions within the research team before being finalised. As part of this process the second and third authors were independently given copies of the Nvivo file and were asked to review the final themes alongside the extracts that comprised each theme. The Nvivo image below shows the number of articles (sources) comprising each theme along with the number of individual references (data extracts).



ıme	Sources	Referen...
strength through adversity	11	43
navigating the system	11	59
living with uncertainty	9	39
connecting with other families	10	29
burden of care	12	45

5. Agreeing theme labels & (6) producing the final report: As a result of the theme review conducted by the second and third authors, it was agreed that the theme labels (produced at stage 3) were credible. Hence no changes were made to the names of the themes at this stage. Finally, the primary author completed a write up of the identified themes, which included use of the CASP tool to critically examine the robustness of the synthesis (Popay et al., 2006).

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Appendix F: Service User Involvement (feedback from the children's hospice charity)

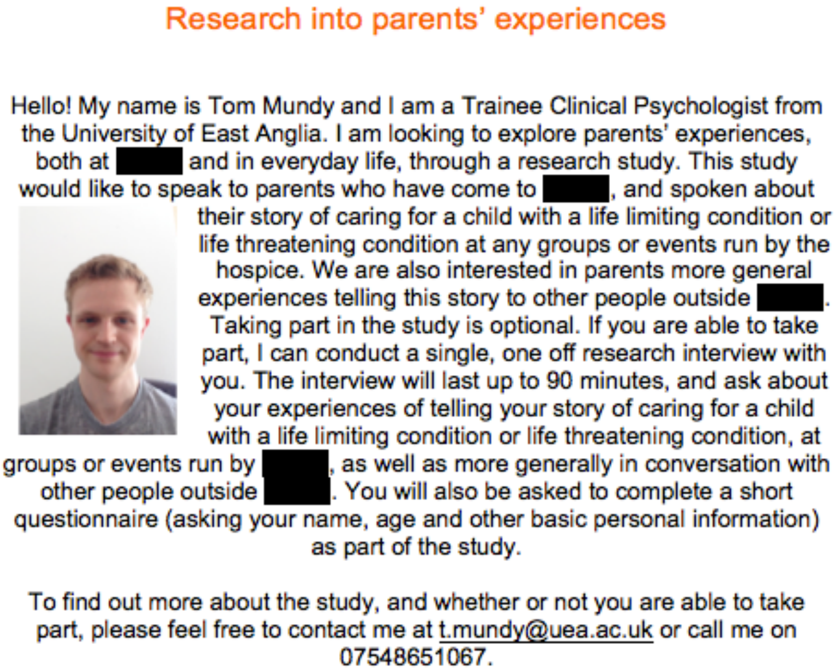
Parents' at the children's hospice charity were asked to review the recruitment materials (the consent form, the information sheet, recruitment flyer) and additional study documentation (interview questions, inclusion and exclusion criteria) relating to the project. Amendments to the documentation have been made following their feedback to ensure appropriate input from service users during the study development phase. This content (i.e. their quoted feedback) has been redacted for the purposes of maintaining confidentiality/anonymity.

Appendix G: Service User Involvement (national PPC organisation)

The recruitment materials (the consent form, the information sheet, recruitment flyer) and additional study documentation (interview questions, inclusion and exclusion criteria) were also reviewed by parents at a separate national PPC organisation supporting parents of children with LLCs. Changes were made following the feedback they provided. This content (i.e. their quoted feedback) has been redacted for the purposes of maintaining confidentiality/anonymity.

Appendix H: Recruitment materials

Newsletter advert



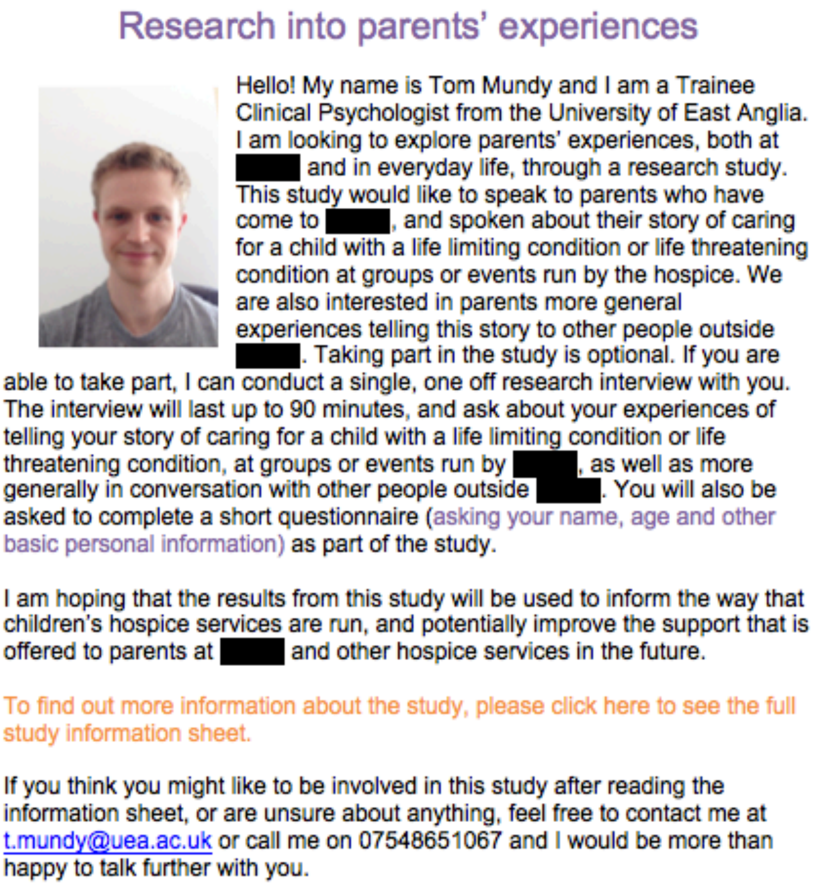
Research into parents' experiences

Hello! My name is Tom Mundy and I am a Trainee Clinical Psychologist from the University of East Anglia. I am looking to explore parents' experiences, both at [redacted] and in everyday life, through a research study. This study would like to speak to parents who have come to [redacted], and spoken about their story of caring for a child with a life limiting condition or life threatening condition at any groups or events run by the hospice. We are also interested in parents more general experiences telling this story to other people outside [redacted]. Taking part in the study is optional. If you are able to take part, I can conduct a single, one off research interview with you. The interview will last up to 90 minutes, and ask about your experiences of telling your story of caring for a child with a life limiting condition or life threatening condition, at groups or events run by [redacted], as well as more generally in conversation with other people outside [redacted]. You will also be asked to complete a short questionnaire (asking your name, age and other basic personal information) as part of the study.

To find out more about the study, and whether or not you are able to take part, please feel free to contact me at t.mundy@uea.ac.uk or call me on 07548651067.

Appendix I: Recruitment Materials

Online advert



Research into parents' experiences

Hello! My name is Tom Mundy and I am a Trainee Clinical Psychologist from the University of East Anglia. I am looking to explore parents' experiences, both at [redacted] and in everyday life, through a research study. This study would like to speak to parents who have come to [redacted], and spoken about their story of caring for a child with a life limiting condition or life threatening condition at groups or events run by the hospice. We are also interested in parents more general experiences telling this story to other people outside [redacted]. Taking part in the study is optional. If you are able to take part, I can conduct a single, one off research interview with you. The interview will last up to 90 minutes, and ask about your experiences of telling your story of caring for a child with a life limiting condition or life threatening condition, at groups or events run by [redacted], as well as more generally in conversation with other people outside [redacted]. You will also be asked to complete a short questionnaire (asking your name, age and other basic personal information) as part of the study.

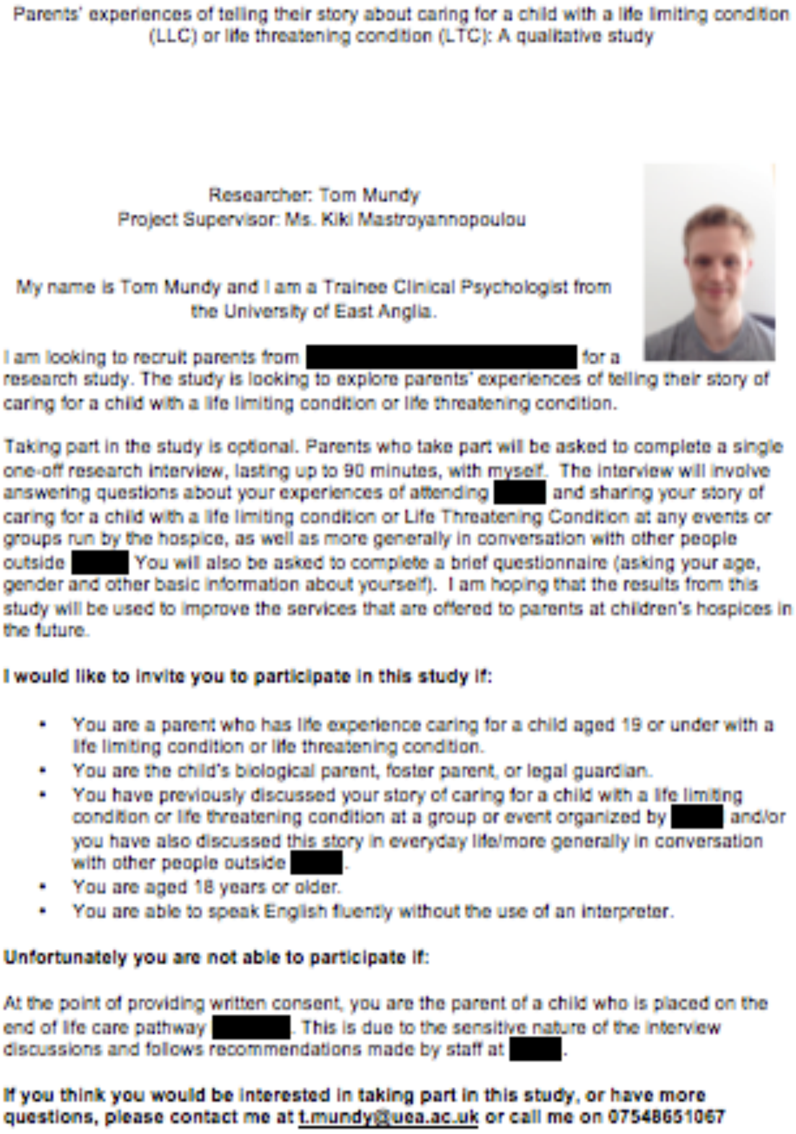
I am hoping that the results from this study will be used to inform the way that children's hospice services are run, and potentially improve the support that is offered to parents at [redacted] and other hospice services in the future.

To find out more information about the study, please click here to see the full study information sheet.

If you think you might like to be involved in this study after reading the information sheet, or are unsure about anything, feel free to contact me at t.mundy@uea.ac.uk or call me on 07548651067 and I would be more than happy to talk further with you.

Appendix J: Recruitment Materials

Flyer



Parents' experiences of telling their story about caring for a child with a life limiting condition (LLC) or life threatening condition (LTC): A qualitative study

Researcher: Tom Mundy
Project Supervisor: Ms. Kiki Mastroyannopoulou

My name is Tom Mundy and I am a Trainee Clinical Psychologist from the University of East Anglia.

I am looking to recruit parents from [redacted] for a research study. The study is looking to explore parents' experiences of telling their story of caring for a child with a life limiting condition or life threatening condition.

Taking part in the study is optional. Parents who take part will be asked to complete a single one-off research interview, lasting up to 90 minutes, with myself. The interview will involve answering questions about your experiences of attending [redacted] and sharing your story of caring for a child with a life limiting condition or Life Threatening Condition at any events or groups run by the hospice, as well as more generally in conversation with other people outside [redacted] You will also be asked to complete a brief questionnaire (asking your age, gender and other basic information about yourself). I am hoping that the results from this study will be used to improve the services that are offered to parents at children's hospices in the future.

I would like to invite you to participate in this study if:

- You are a parent who has life experience caring for a child aged 19 or under with a life limiting condition or life threatening condition.
- You are the child's biological parent, foster parent, or legal guardian.
- You have previously discussed your story of caring for a child with a life limiting condition or life threatening condition at a group or event organized by [redacted] and/or you have also discussed this story in everyday life/more generally in conversation with other people outside [redacted].
- You are aged 18 years or older.
- You are able to speak English fluently without the use of an interpreter.

Unfortunately you are not able to participate if:

At the point of providing written consent, you are the parent of a child who is placed on the end of life care pathway [redacted]. This is due to the sensitive nature of the interview discussions and follows recommendations made by staff at [redacted].

If you think you would be interested in taking part in this study, or have more questions, please contact me at t.mundy@uea.ac.uk or call me on 07548651067

Appendix K: FMH Research Ethical Approval

Floor 1, The Registry
University of East Anglia
Norwich Research Park
Norwich, NR4 7TJ

Email: fmh.ethics@uea.ac.uk

Web: www.uea.ac.uk/researchandenterprise

Thomas Mundy
MED

23.10.17

Dear Thomas,

Title: Parents' experiences of telling their story about caring for a child with a life limiting condition (LLC) or life threatening condition (LTC): A qualitative research study

Reference: 2017/8 - 11

The submission of your above proposal has been considered by the Faculty Research Ethics Committee and we can confirm that your proposal has been approved.

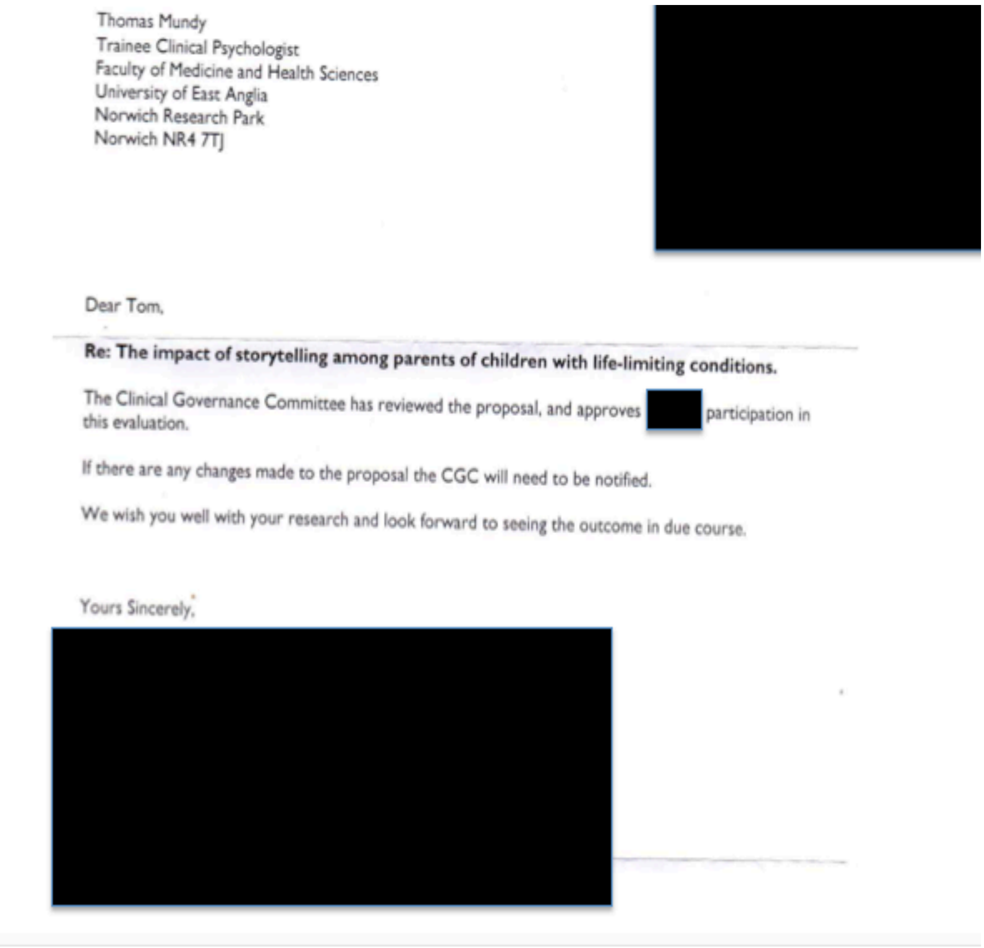
Please could you ensure that any further amendments to either the protocol or documents submitted are notified to us in advance and also that any adverse events which occur during your project are reported to the Committee. Please could you also arrange to send us a report once your project is completed.

Yours sincerely,

[redacted]

CC Kiki Mastroyannopoulou

Appendix L: Children's Hospice Charity Ethical Approval



Thomas Mundy
Trainee Clinical Psychologist
Faculty of Medicine and Health Sciences
University of East Anglia
Norwich Research Park
Norwich NR4 7TJ

[redacted]

Dear Tom,

Re: The impact of storytelling among parents of children with life-limiting conditions.

The Clinical Governance Committee has reviewed the proposal, and approves [redacted] participation in this evaluation.

If there are any changes made to the proposal the CGC will need to be notified.

We wish you well with your research and look forward to seeing the outcome in due course.

Yours Sincerely,

[redacted]

The 'RE title' above is an abbreviated version of the full title, but refers to the study implemented in this thesis; the children's hospice charity were provided with the full approved UEA FMH approved protocol/proposal (including UEA FMH ethics approval letter) in order to reach their decision, which this letter refers to. The study adhered to the proposal referred to at all times. Note: the approval attached entailed ethical approval for all 3 recruitment sites owned by the children's hospice charity. Senior staff at the hospice charity (including the study collaborator/head of the clinical governance committee) (as well as the head of UEA FMH ethics & three UEA ClinPsyD staff members) agreed that the letter above provided adequate evidence of the hospice charity providing ethical approval for the project described within this thesis. The hospice charity were also made aware & accepted that the study title used in the thesis write up differed marginally from this letter.

Appendix M: Consent Form

Version 1.

Parents' experiences of telling their story about caring for a child with a life limiting condition (LLC) or life threatening condition (LTC): A qualitative research study

Researcher: Tom Mundy
Supervisor: Ms. Kiki Mastroyannopoulou
Study Collaborator: [redacted]

Please initial each box

1. I confirm that I have read and understood the study information sheet dated.................... (version............) for the above study. ☐

2. I have had the opportunity to ask questions about the research study and have had all my questions answered satisfactorily. ☐

3. I understand that my participation is voluntary and that I have a right to withdraw from the study at any time, without giving any reason, up until two weeks after the interview has been completed. ☐

4. I agree to take part in a research interview lasting approximately 90 minutes. ☐

5. I understand that anonymised quotes or extracts from the interview may be included within the final report. ☐

6. I consent to the interview being recorded using a dictaphone, ☐

7. I understand that involvement in a research interview for the study will not have any impact on the care or treatment any member of my family (including myself) receive from [redacted] ☐

8. I understand that any information I provide is confidential, and my information will be stored securely in accordance with UEA Data Management Policy (2015) and the Data Protection Act (1998). ☐

I would like to receive a copy of the completed interview transcript, so that I can provide feedback and comment upon its accuracy (please tick the appropriate box):

YES ☐ NO ☐

I would like to receive a copy of the final study report (please tick the appropriate box):

YES ☐ NO ☐

Participant:

__________________ __________________
Name of Participant Signature

Date:

Researcher:

__________________ __________________
Name of Researcher Signature

Date:

Appendix N: Information Sheet

Study Information Sheet
Version 1.

Participant Information Sheet

Parents' experiences of telling their story about caring for a child with a life limiting condition (LLC) or life threatening condition (LTC): A qualitative research study

Researcher: Tom Mundy
UEA Project Supervisor: Ms. Kiki Mastroyannopoulou
Study Collaborator: [redacted]

Caring for a child with a life limiting condition or life threatening condition can be a difficult experience for many parents. In order to improve the support parents receive and ensure they get the best possible help from children's hospice services, we are looking to recruit parents from [redacted] for a research study. The study is looking to investigate parents' experiences of telling their story of caring for a child with a life limiting condition or life threatening condition.

Taking part in the study is optional. Before deciding whether or not you would like to take part, take the time to read through this information sheet carefully. If you have any questions about this research study, or think you might like to participate, the researchers contact details can be found at the end of the information sheet.

We would like to invite you to participate in this study if:

- You are a parent who has life experience caring for a child (aged 19 or under) with a life limiting condition or life threatening condition.
- You are the child's biological parent, foster parent, or legal guardian.
- You have previously discussed your story of caring for a child with a life limiting condition or life threatening condition at a group or event organized by [redacted] and/or you have also discussed this story in everyday life/more generally in conversation with other people outside [redacted]
- You are aged 18 years or older.
- You are able to speak English fluently without the use of an interpreter.

Unfortunately you are not able to participate if:

- At the point of providing written consent, you are the parent of a child who is placed on the end of life care pathway by [redacted] This is due to the sensitive nature of the interview discussions and follows recommendations made by staff at [redacted]

What is the purpose of this study?

This study is looking to explore parents' experiences of telling their story of caring for a child with a life limiting condition or life threatening condition. The study is interested in any experiences parents' may have had, either good or bad, about telling this story at a group or event run by [redacted] This could be any group or event, including (but not limited to) parties, supportive groups, fundraising events, or choirs organized by [redacted] We also interested in parents' more general experiences telling this story in conversation with other people outside [redacted] The project is looking to learn more about parents' experiences in this area, so that hopefully ways can be found to improve the services that are offered to parents at children's hospices in the future.

The research study is being conducted by Tom Mundy, a Trainee Clinical Psychologist at the University of East Anglia (UEA), as part of his Doctorate in Clinical Psychology degree. . The research project has been organized jointly by UEA and [redacted] A clinical lecturer at UEA, Ms. Kiki Mastroyannopoulou, is the primary project supervisor[redacted] , is also supporting the research project.

Do I have to participate?

No. Participating in this study is voluntary. It is entirely up to you whether or not you decide to take part. If you decide not to participate, this will have no impact on you in any way. You will also be able to withdraw from the study, without giving any reason, up until two weeks after the interview has been completed.

What will happen if I take part?

If you are eligible to participate the researcher will arrange a time and date to meet with you in person for a research interview. To limit the demands on your time, the interview can take place at the hospice or at your home[9], depending on which location you prefer. If finding childcare is a problem, the researcher is also able to be flexible with timings and interview dates to help accommodate your needs.

We also appreciate that you may have worries or concerns before the interview starts. The researcher will be able to talk to you again in person before the interview begins, and take the time to answer any remaining questions you have. If you agree to take part, he will then ask you to sign a consent form and complete a single research interview, which will last approximately 90 minutes. In order to enable yourself and the researcher to focus, the interviews will ideally be conducted one to one, without any other individuals present. In the event that you would like someone else to be present with you during the interview (e.g. your child), please discuss this with the researcher prior to the interview. The interview itself will involve answering questions about your experiences of sharing your story of caring for a child with a life limiting condition or life threatening condition. During the interview you will also be asked to complete a basic questionnaire, asking your age, gender, ethnicity, educational status and employment level. The interview will be audio recorded using a Dictaphone.

---

[9] Potential existed for interviews to be undertaken outside of these locations, where ethical and appropriate, in accordance with the study protocol (e.g. the workplace).

If you choose to have the interview completed at your home address, basic personal details (your name and home address) will be shared with a nominated UEA colleague [redacted] and noted down by this individual immediately prior to the interview visit. This information is shared only for lone working purposes. After the interview has ended, the researcher will call the nominated UEA colleague, and ensure that the copy of your personal information held by this individual is immediately destroyed. You will not be asked to complete any additional research interviews or speak publicly about your experiences as part of the study.

How will the information from the interviews be used?

Once the interview has been completed, the researcher will type out the interview word for word to create a written transcript (a transcript is a written record of what was said during the interview). During transcription the researcher will anonymise the interview data by altering or removing any personal information. This will involve giving you and any other individuals you mention a pseudonym (a name different from your real one). The researcher will ask you for the pseudonyms you would like to use during the interview. With your consent, the researcher will send the anonymised transcript to you, so that you can check the interview transcript and ensure it is accurate.

The researcher will then analyze the interview transcript to identify any important themes using an approach called Interpretative Phenomenological Analysis (IPA). The researcher will discuss extracts from the interview transcripts with the primary supervisor (Ms. Kiki Mastroyannopoulou) to ensure the themes are carefully examined for their accuracy.

After the interviews have been analyzed, the researcher will write a report describing the main findings from the interviews. In the report, quotes and extracts from the interviews will be used to illustrate key themes. However, the researcher will not include any information that could personally identify you, or any other individual you mention during the interview.

After the report is written it will be submitted to the University of East Anglia. Staff from the University and external examiners will view the report, and it may be selected for publication in a scientific journal. The reports findings will also be given to staff at [redacted] However you will not be not be personally identified at any time in the report. You will also be given the option to receive a copy of the final report once it has been completed.

How will my information be stored?

The conversation during the interview will be recorded on a Dictaphone. After the interview the researcher will store this information securely on an encrypted USB memory stick. The researcher will then delete the audio recording from the Dictaphone. The transcript will be saved securely on the encrypted USB memory stick mentioned above. A further copy of the audio recording and interview transcript will be stored on a password-protected computer based at the University of East Anglia.

All consent forms and other information with your personal details will be stored securely at the University of East Anglia, Elizabeth Fry Building, in a locked file draw. Only the researcher and research supervisor will have access to any information you provide. All information will be stored securely for up to 10 years in keeping with the University of East Anglia Data Management Policy (2015), before being destroyed.

Are there any advantages if I take part?

The findings will contribute to our understanding of what it is like for parents to talk with other people about caring for a child with a life limiting condition or life threatening condition. The researcher hopes that the results from this study will be used to improve the support offered to parents at children's hospice services in the future.

Are there any risks or disadvantages if I take part in the study?

It is unlikely that this study will pose any risk to your safety or wellbeing. However if at any point you begin to feel upset, you will be able to end the interview, or take a break.

If you disclose information during the interview that makes the researcher concerned about your safety, or the safety of someone else, he will be obliged to contact [redacted] [redacted] and the primary project supervisor, Ms. Kiki Mastroyannopoulou.In the event that further action is required (or it is not possible to contact [redacted] the researcher may also need to contact the duty manager at [redacted] or other relevant third parties to ensure that any risks are properly managed.

If I am unhappy about this research project – how do I make a complaint?

Complaints can be made to [redacted]

I am interested in taking part– how can I find out more information?

If you have any further questions about this study, or think you might be interested in participating, you can contact the researcher, Tom Mundy, via email at t.mundy@uea.ac.uk or call him on 07548651067.

Researcher Contact Details:

Tom Mundy,
Trainee Clinical Psychologist,
Email: t.mundy@uea.ac.uk
Tel: 07548651067

Supervisor Contact Details:

Ms. Kiki Mastroyannopoulou
Clinical Lecturer
Email:
k.mastroyannopoulou@uea.ac.uk
Tel: 01603 59 **3961**

Appendix O: Demographic Questionnaire

Version 1.

This questionnaire asks you for some basic demographic information. If you would prefer not to give an answer, please tick the option ‘not disclosed’.

What is your gender? (Please tick the appropriate option.)

- Male
- Female
- Other (Please Specify):
- Not Disclosed

Which age group do you fall into? (Please tick the appropriate option.)

- 18 to 24 years
- 25 to 34 years
- 35 to 44 years
- 45 to 54 years
- 55 to 64 years
- Age 65 or older
- Not disclosed

What is your race/ethnicity? (Please tick the appropriate option.)

- White British/ White Irish / White Other
- Mixed/ Multiple Ethnic Groups
- Black/ African / Caribbean / Black British

- Asian / Asian British
- Other (Please Specify):
- Not Disclosed

Employment status (please tick the appropriate option.)

- Employed full time (30 or more hours per week)
- Employed part time
- Unemployed
- Full time student
- Retired
- Full time carer
- Other (Please Specify):
- Not disclosed

Educational level (please tick the highest level of educational attainment you have.)

- No educational qualifications
- GCSE/O-Level
- AS/A-Levels
- University undergraduate degree
- University postgraduate degree
- Other higher education or professional qualification. Please state:
- Not disclosed

Appendix P: Interview Topic Guide

Topic Guide

Following service user recommendations each interview began with more general descriptive questions to put the parent at ease: How long have you been coming to the hospice? When did you first attend? How old is your child? Would you feel able to tell me what their diagnosis is?

Primary Questions

1. What sort of things of are you aware of when you are talking about your story of caring for a child with a LLC or LTC at a group or event run by the hospice?

2. What are the positive aspects of telling your story?

3. What are the difficult aspects of telling your story?

4. Is your experience of telling your story different depending on audience and in what way?

5. Has your experience of telling your story changed over time or evolved?

6. Think of a specific experience of telling your story in conversation with other people outside the hospice.. What was it like to tell the story on this occasion?

Appendix Q: Post Interview Information Sheet

Thank you for taking part in this research study. If you have any further questions about the research after the interview has been completed, you can contact the researcher, Tom Mundy via t.mundy@uea.ac.uk or by calling 07548651067.

We appreciate that you have been asked to talk about emotive topics during the research interview. If for any reason this interview has been upsetting, and you would like to receive further support, please contact the appropriate services that are listed below.

[redacted]

The [redacted] can meet with you to provide further support for your emotional health and wellbeing. A self-referral can be made directly by calling one of the numbers below and asking to speak to a member of [redacted] They are available 9am-4pm, Monday-Friday.

If you live in [redacted] can call [redacted]

If you live in [redacted] you can call [redacted]

If you live in [redacted] you can call [redacted]

[redacted] Services

If you are living in [redacted] you can call the [redacted] Service on [redacted] (8am until 8pm, Monday-Friday). They can provide advice and assessment regarding any mental health difficulties you may be experiencing.

If you are living in [redacted] you can call the [redacted] Service. This is a 24-hour service for people in a mental health crisis. **Call [redacted] and press [redacted] to access the service. Specially trained staff will be able to speak with you and discuss any needs you have.**

If you need general medical help or advice, you can also call the [redacted] service free of charge on telephone number [redacted] The service can also direct you if you're not sure which [redacted] services you need.

Alternatively you can make an appointment to see your GP at your nearest surgery.

Other Useful Contacts

[redacted] This service provides information and support for parents of children and young people that are expected to have a short life. The [redacted] Helpline can be called free on [redacted] [redacted] They can also be emailed at [redacted]

[redacted] is a confidential listening service staffed by trained volunteers. The service is available 24 hours a day, 365 days a year on the telephone number [redacted] The telephone number is free to call. You can also email the [redacted]

[redacted] Advice Line: This service provides information and advice regarding mental health issues. You can call the advice line on [redacted] (10am-2pm Monday to Friday) or email them at [redacted]

In An Emergency

If you, or anyone else you know is in immediate danger, please call 999 or go to your closest A&E department.

Appendix R: Risk Management Protocol

Note: The Protocol below was formally approved by the hospice charity [redacted] and UEA as part of the ethics process

Participant wellbeing

The study will not involve any form of deception and no financial inducements will be provided in return for participation. Each participant will be fully informed about all aspects of the research project in order to ensure informed consent is obtained.

The evidence base indicates that studies of this type are unlikely to cause significant distress (e.g. Stevens et al., 2003). However, in the event that the parent experiences any distress the researcher will offer additional breaks and remind the participant that they can postpone or terminate the interview at any time. An information sheet with sources of support, including the contact telephone numbers for various mental health services, will be offered to all participants at the interview exit stage. Within the information sheet every parent will be given contact information for the [redacted] at [redacted] and informed that they are able to access this service via telephone self-referral.

Where risks involving harm to the parent or another person are identified the interviewer will follow a risk management plan. Implementing the plan will involve undertaking a brief examination of the problem to obtain further information about risk severity. The researcher will then contact [redacted] by telephone on the same day to discuss the issue and ensure appropriate action is taken. In the event that [redacted] cannot be contacted the on-site duty manager will be contacted by telephone on the same day at [redacted] The supervisor will then be informed as soon as possible to ensure appropriate action has been taken to manage risk.

Researcher wellbeing

As the project involves potential exposure to distressing emotional content the researchers wellbeing will be supported through additional supervisory input. The researcher will meet with the primary supervisor for extra one-to-one supervision on a monthly basis to debrief and explore any difficult experiences after each interview. Should the researcher require further support, this will be discussed with the primary supervisor to ensure further appropriate input is obtained.

In order to critically appraise the impact of ones personal actions on the participants and the research process, the researcher will keep a reflective diary throughout the empirical project. Written comments will be used to supplement the interview content by documenting relevant themes, non-verbal features of the participant's behaviour as well as the way that the researchers own values, preconceptions, and conduct may influence parents' narratives or affect interpretations of their responses (Nadin & Cassell, 2006; Jootun, Mcghee, & Marland, 2009). Extracts from the journal dealing with these issues will inform discussions within supervision, as this will allow the researcher to develop a more detailed, insightful analysis, based on a reflexive position (Braun & Clarke, 2003). No identifying participant information will be included within the reflective diary.

The researcher will give adequate consideration to his personal safety. Interviews will be completed in the working day (9am-5pm) where possible. A study mobile phone will allow the researcher to call others for emergency assistance if required, or verify the researchers safety if staff have concerns about his wellbeing. Where home visits are conducted the researcher will maintain contact with a nominated colleague [redacted] by calling this individual before and after each appointment (UEA, 2013). The location and the expected length of the visit will be given to the nominated colleague immediately before the interview. Following the interview, the researcher will contact the nominated colleague and ensure the participants personal details are immediately destroyed. In the event that the researcher fails to make contact at the anticipated time, the nominated colleague will attempt to call the study mobile phone, before contacting the primary supervisor to ensure appropriate further action is taken. These actions are necessary in order to ensure compliance with the UEA lone worker policy (UEA, 2013) and the lone worker policy at [redacted]

Appendix S: Coding Extracts

Participant 1

Interviewer; Because there must have been a point where you first came into the hospice and would have had to tell them about, the story I suppose you have just related to me.

Participant: Yeah

Interviewer: And do you remember what that experience was like?

Participant: The lady (counsellor) that I am seeing now, is well, is, is, great because she, (pauses) she’s very empathetic, and I sort of tell my story with the recognition that there are children at the hospice that are much more severe, or sort of end of life, so trying to sort of bear that in mind, and think my experience has probably not been as traumatizing as theirs has. Um but she’s very good at sort of acknowledging the difficulties, and also like how atypical my experience was.

The counselor provided empathy and a sense of understanding, but the parent seems to contrast this with a sense that it is important to remember that other people are also worse off than her. She places her situation in a much wider context by doing this.

The parent feels she has been understood by the therapist at the hospice, the therapist is able to appreciate the difficulties she has encountered.

The parent needs someone to appreciate the uniqueness of her situation and validate this in an empathic way.

The parent is engaged in an act of social comparison against others, possibly this leads her to minimize her difficulties.

Participant 2

Interviewer: and do you remember telling your story at those groups and what that experience was like? Participant: Yeah yeah, over time. There were 2 families in particular that we met through the group who um we are friends with now, um we don't see each other a lot because we've all got the same difficulties children with complex needs, but yeah I remember like a few times talking to them and I think for all of us we'd, we'd not got anyone else in that sort of boat, no one else to talk about our fears for the future and the day to day of living with it and I think for all of us it was so important, and I've always thought that even if you can just find one other person in your boat it makes such a massive difference. For me, we've become friends with another family, who we met at the hospice through a local group and are still good friends now, and I remember getting to know them took away a lot of our self pity, because when you were in one of those crazy situations, (pause) instead of just going, I cant believe this is happening or whatever, it was more like, oh it happens to them too and it was you just didn't feel like you were the only ones.

The parent seems to have felt isolated prior to telling her story. Implicitly it feels there is a lack of understanding from the wider community regarding the situation she finds herself in.

Exchanging stories has a cathartic dimension to it. Parents are able to bond over the commonality of their experience. It also seems that this is emotionally helpful for the parent during the more turbulent and challenging times of the child's illness.

Parents formed new relationships with one another through storytelling, there is a possible feeling of closeness to others in the same situation.

The parent is building up a support network. Not feeling so isolated or alone is implicit.

Participant 4

The parent feels a rapport with others in a similar situation to herself.

Similarity of condition and trajectory of illness is influential in the experience – parent sometimes feels wary when telling her story?

Interviewer: So does that mean you sometimes hold back what you say in your story or there are details you leave out?

Participant: I think it depends on who you tell the story to. I think if it's someone similar to you then I think you just let it all out and its easy to let it out. If they are not then you can't do, especially if their child is not as disabled. Or their child is worse than yours. I think it also depends on your own mood. You know if I'm having a rough time I cant be bothered to talk about it again and again and then there are other times when we cant stop talking about it. Because however hard it is to hear it in your own head sometimes its good to talk about it and take the pressure off yourself I think.

Seems to provide evidence that the experience varies.

There is a cathartic, or therapeutic dimension to telling the story.

Participant 5

You know you kind of explain it for half an hour, before they are even thinking of how to treat your daughter. It should all be on the file, they should just be able to do what they need to do, then talk about it. But I think a lot of parents go through the same thing, so its almost like you want to have it written down, and then, each time you go to someone new or you go to…a bit like your information sheet you can just give it to them, say there it is.

The parent appears to feel let down by services in this particular instance.

The parent is contrasting his desire for a better system with the need to go over the story continuously. Here the telling of the story feels monotonous.

Writing down the story could be useful for others in the same situation.

Participant 6

Yeah. I suppose the difference between therapeutically telling the story and having to kind of say it or not so much I guess its more now, just thinking about it now that, we've been there for a while you having to keep doing it to get things, get things to happen, er, I don't know if that makes sense, to get that support, whereas I guess maybe at the start. I hadn't really thought about it this much before. You know at the start, at the first time in the hospice or during that period, I think part of it was er, like being a therapeutic I guess, that this is good to talk about it, that kind of thing, whereas, id say now its changed to having to do it again and again and again and again.

The repetition of the story feels anti-therapeutic here.

Telling the story is helpful or beneficial to the parent, talking about it is initially seen as inherently good. It helped to tell the story at the hospice.

Telling the story is possibly a means of advocating for the child and their needs.

Participant 8

Interviewer: And did you get an opportunity to tell the story to those parents, what was that like?

Participant: The hospice, in the hospice, it was so nice, it was so relaxed. So, and you know that in there, other parents they will have similar situations, like my family. So I, as I remember I didn't have any difficulty to tell anyone, or think about it, or think should I say that? Should I not say that? I cannot remember this experience. We sit in the room, in the room there and had a bit of a chat and say our story. So if you know someone has a similar situation, there is no problem to say about it at all. I think I would really like to share a little bit more about my daughter, not try to hide something, I would just like to share everything.

The parent tells the story with apparent ease to likeminded individuals. She does not appear as guarded within the hospice context as in the outside world.

She does not appear to be monitoring the story, she is possibly on the lookout to find someone in a similar situation to herself?

The hospice is perceived as being a place of safety for the parent, as distinct from the outside world? The story develops and emerges through the parents own self monitoring by asking questions.

It sounds as if there are few opportunities for her to tell the story in an open way. Possibly looking to tell the story in order to give the child more of a voice?