The Journey through Care

Palliative Care for children and young people, and their families: What? When? How?

A Realist Inquiry
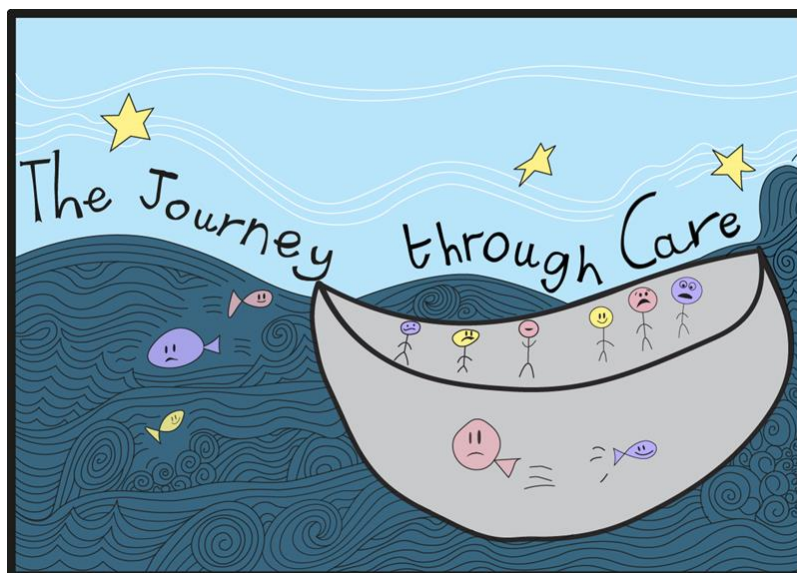
Dr Sarah Jane Mitchell
MBChB FRCGP MSc Pall Med

Supervisors: Professor Jeremy Dale, Professor Anne-Marie Slowther,
Professor Jane Coad

Thesis submitted for fulfilment of the requirements for the Degree of
Doctor of Philosophy in Health Sciences

Warwick Medical School
University of Warwick
February 2020

Table of Contents

Tables

Boxes

Photographs

Figures

Disclaimer

Acknowledgements

Declaration

This thesis is submitted to the University of Warwick in support of my application for the degree of Doctor of Philosophy. It has been composed by myself and has not been submitted in any previous application for any degree.

I have carried out the work presented (including data generated and data analysis) in this thesis, supported by a research team comprising my supervisors unless otherwise stated. Transcription of interviews and focus groups was carried out both by me, and a university approved transcription service. Warwick medical students, Karina Bennett and Andrew Morris, took part in data extraction and synthesis for the literature reviews. Permission has been granted from all individuals in the photos to use them in this thesis. No participants feature in any of the photos, those who do are all people I know personally. The comic strips slides presented in Appendix 8 were created using Storyboardthat.com. A subscription has been paid. The company retains the rights to the illustrations.

Parts of this thesis that have been published are as follows:

- Mitchell S, Slowther A, Coad J, Dale J. The journey through care: study protocol for a longitudinal qualitative interview study to investigate the healthcare experiences and preferences of children and young people with life-limiting and life-threatening conditions and their families in the West Midlands, UK. *BMJ Open* 2018;**8:**e018266. doi: 10.1136/bmjopen-2017-018266
- Mitchell S, Slowther A-M, Coad J, Akhtar S, Hyde E, Khan D, Dale J. Ethics and Patient and Public Involvement with Children and Young People. *Arch Dis Child Ed Pract*, Published Online First: 09 February 2018. doi: 10.1136/archdischild-2017-313480
- Mitchell S, Morris A, Bennett K, Sajid L, Dale J. "Specialist paediatric palliative care services: what are the benefits?" *Arch Dis Child* Published Online First: 04 April 2017. doi: 10.1136/archdischild-2016-312026
- Mitchell S, Bennett K, Morris A, Slowther A-M, Coad J, & Dale, J. (2019). Achieving beneficial outcomes for children with life-limiting and life-threatening conditions receiving palliative care and their families: A realist review. *Palliat Med*. doi.org/10.1177/0269216319870647

Signed: Date: 8th December 2019

Abstract

Background:
The World Health Organisation defines palliative care as "an approach to care that aims to improve the quality of life for people living facing the problems associated with life-threatening illness, through the prevention and relief of suffering". The number of children and young people with life-limiting and life-threatening conditions is rising. All could potentially benefit from palliative care, but there are significant inequalities in the provision of such care to children internationally, including the availability of specialist paediatric palliative care services.

Research Aims:
To examine the delivery of healthcare, including palliative care, for children with life-limiting and life-threatening conditions and their families. The study uses a realist approach to provide understanding into how palliative care is delivered most effectively, and when, leading to policy relevant recommendations.

Methods:
1. Development of a programme theory (PT) through systematic and realist literature reviews.
2. 41 serial interviews with 31 participants from 14 families; 10 children with life-limiting or life-threatening conditions and 21 family members.
3. Four focus groups with children's palliative care professionals.
4. Thematic / realist analysis to describe the hidden mechanisms (M), triggered in certain contexts (C), to produce desired outcomes (O). Context-Mechanism-Outcome configurations (CMOCs) are used to refine and refute the PT.

Findings:
Children and their families are vulnerable experts, negotiating a healthcare system that can be rigid and fragmented (C). The delivery of palliative care depends on interpersonal relationships with healthcare professionals (C), who are able to bear witness to the child and family situation (M), underpinned by trust and respect (M). Important child and family outcomes include feeling heard and supported (O). In organisations, an environment that values these relationships and legitimises palliative care as an approach (C) through leadership and role modelling (M) as well as the development of specialist services (C), leads to more equitable palliative care (O).

Conclusion:
The realist approach provides increased understanding and description of important child and family outcomes that underpin policy goals in palliative care, achieved in certain contexts. Future service and commissioning models should propose a whole system approach. Achieving this requires the unwavering commitment of system leaders, recognition of the unique situations of children and families, and individualised palliative care. Service design should place greater emphasis on the need for trusted relationships, should nurture and support professionals who have the motivation and capacity to provide palliative care, and should integrate specialist paediatric palliative care effectively into existing services.

Plain English Summary

"Palliative care" is an approach to care that can improve the quality of life of children with a life-limiting or life-threatening condition (incurable conditions, or serious conditions that might be cured). The number of children living with life-limiting and life-threatening conditions is rising, and there is a need for more evidence to understand when and how palliative care can benefit these children and their families.

The aim of this research was to provide an increased understanding of how children with life-limiting or life-threatening conditions, and their family members, experience the current healthcare system, and the effect this has on whether or not they experience palliative care. The research used realist methods to understand how palliative care is delivered most effectively, to which children, and when. Patient and public involvement with young people played an important in the design of the study.

The research started with literature reviews (looking at research and policy documents). These were followed by an interview study with 14 families (ten children with life-limiting or life-threatening conditions and 21 of their family members took part). Each family took part in up to three interviews over 13 months. Healthcare professionals working in a range of children's healthcare services, including children's hospices and specialist paediatric palliative care in hospitals took part in four focus groups.

Findings showed that children and families want to feel respected, heard and supported by their healthcare services as they face difficult situations, such as frequent deteriorations in their child's condition. These situations can be emotionally demanding for healthcare professionals too, and the delivery of palliative care depends on the development of trusted relationships between the child, their family, and their healthcare professionals. Senior leaders in healthcare have a role in promoting a palliative care approach and changing culture within healthcare organisations.

Abbreviations

ACP   Advance Care Plan

AHP   Allied Healthcare Practitioner

A&E   Accident and Emergency

BCH   Birmingham Children's Hospital

BNFC   British National Formulary for Children

CCG   Clinical Commissioning Group

COPD   Chronic Obstructive Pulmonary Disease

CMOC   Context-Mechanism-Outcome Configuration

CRD   Centre for Reviews and Dissemination

CRN   Clinical Research Network

CT   Computerised tomography

DRF   Doctoral Research Fellowship

EBM   Evidence Based Medicine

ECMO   Extracorporeal Membrane Oxygenation

ED   Emergency Department

GP   General Practitioner

GRIPP   Guidance for Reporting Involvement of Patients and the Public

HRA   Health Research Authority

ICPCN   International Children's Palliative Care Network

ICU   Intensive Care Unit

LTV   Long Term Ventilation

MCA   Mental Capacity Act

MDT   Multi-Disciplinary Team

NGT   Naso-gastric Tube

NHS   National Health Service

NICE   National Institute for Health and Care Excellence

NIHR   National Institute for Health Research

NPT   Normalisation Process Theory

PCA   Patient Controlled Analgesia

PEG   Percutaneous Endoscopic Gastrostomy

PIC   Paediatric Intensive Care

PICOS   Patient, problem or population, Intervention, Comparison, Outcome

PICU   Paediatric Intensive Care Unit

PPI   Patient and Public Involvement

PRISMA Preferred Reporting Items for Systematic Reviews and Meta-Analyses

RCGP   Royal College of General Practitioners

RCPCH Royal College of Paediatrics and Child Health

REC   Research Ethics Committee

SPPC   Specialist Paediatric Palliative Care Service

RCT   Randomised Controlled Trial

TPN   Total Parenteral Nutrition

USA   United States of America

UK   United Kingdom

VBM   Values-based Medicine

WHO   World Health Organisation

YPSG   Young Person's Steering Group

YPAG   Young Person's Advisory Group

Overview of the thesis

The thesis examines "palliative care", defined as an approach that aims to improve the quality of life of anyone, including children, living with a life-limiting or life-threatening condition (1, 2). The thesis has twelve chapters, divided into four parts. Part One provides the background to the thesis. Chapter 1, the introduction starts with the problem statement. The current provision of palliative care for children is described, and the rationale and motivation for the research are outlined. The research questions are then proposed. Patient and public involvement (PPI) informed the design, conduct, analysis and dissemination of the research and is described in Chapter 2. The chapter includes an overview of the ethical approach taken (3) and an evaluation of the experiences of the young people who took part.

Chapter 3 is the literature reviews chapter, which examines the research evidence regarding the effects of palliative care provision on the child and family experience. There is a systematic review, which asks, "what are the benefits of specialist paediatric palliative care?" (4), and a realist review, which provides more in-depth insight into how palliative care works, who for and in what circumstances (5).

Part Two of the thesis outlines the methodology and methods, with the research methodology discussed in chapter 4, and the research methods in chapter 5. The research protocol has been published in an open access journal (6).

The findings of the research are presented in Part Three. Chapter 6 describes the outcomes of the recruitment strategy, and introduces the study population. Chapter 7 presents the findings of the thematic analysis related to the child and family experience of living with a life-limiting or life-threatening condition, and interactions with the healthcare system at an interpersonal (micro-system) level. Chapter 8 describes findings related to the family experience of healthcare organisations and the healthcare system (meso and macro-system levels). A realist logic is applied to the analysis to propose context-mechanism-outcome configurations in Chapter 9.

The final part of the thesis starts with Chapter 10, in which the findings of the empirical research and the literature review are brought together in order to develop and propose the programme theory. Chapter 11 starts with a discussion of the research findings, relating these to the research questions. Policy relevant recommendations are proposed, drawn from the programme theory. The applicability of the programme theory and its implications for policy and practice are discussed. The methodological strengths and limitations of the study are also considered, and the thesis is concluded in Chapter 12.

Part One: Background

1. Introduction

1.1. Overview of Chapter 1

Chapter 1 provides a problem statement, which is the starting point for this thesis. The problem statement is followed by an outline of the current provision of palliative care for children in the UK National Health Service (NHS) and internationally, and an initial programme theory. The chapter includes an explanation of my motivation for the research, the rationale, and the research questions.

Age ranges for children and young people as a population in research and policy vary. For the purposes of this thesis, children and young people have been referred to as "children" throughout, except in Chapter 2 (Patient and Public Involvement), where they are referred to as young people. Age ranges included at each stage of the research (literature reviews and empirical research) are stated and explained in these sections of the thesis.

1.2. Problem Statement

"Palliative care" is advocated in national and international policy as an approach that aims to improve the quality of life of anyone, including children, who is living with a life-limiting or life-threatening condition (1, 2). An increasing number of children are living with life-limiting conditions (those which cannot be cured and which will cause premature death) and life-threatening conditions (where curative treatment is possible but may fail) (7). Their conditions are often fragile and unpredictable, are associated with long hospital stays and the use of medical technology, and change family life forever (8). Having to contemplate the possibility that a child may die is an unbearable and terrifying prospect, against the natural order of events (9, 10), but is a reality for a growing number of families.

Globally, over seven million children (aged 0-19) could benefit from palliative care, with at least 49,000 in the UK, and this number is rising (7, 11). As children live longer with more complex and unpredictable life-limiting and life-threatening conditions, there is an urgent need for research to enhance understanding of when and how palliative care can provide benefit to children and families. This is essential to inform future commissioning models and the design of healthcare services concerned with the provision of palliative care (12).

1.3. Background

Defining palliative care

Palliative care is defined by the World Health Organisation (WHO) as "an approach that improves the quality of life of patients and their families facing the problems associated with life-threatening illness, through the prevention and relief of suffering by means of early identification, assessment and treatment of pain and other problems, physical, psychosocial and spiritual (1)". The WHO expand this definition for children with life-threatening conditions, describing it as "a special, albeit closely related field to adult palliative care". The principles of palliative care which are outlined include:

- The active total care of the child's body, mind and spirit, and support to the family.
- It begins when illness is diagnosed, and continues regardless of whether or not a child receives treatment directed at the disease.
- Health providers must evaluate and alleviate a child's physical, psychological, and social distress.
- Effective palliative care requires a broad multidisciplinary approach that includes the family and makes use of available community resources; it can be successfully implemented even if resources are limited.
- It can be provided in tertiary care facilities, in community health centres and even in children's homes"(1)"

The UK national charity for paediatric palliative care, Together for Short Lives, provide an alternative but widely accepted definition for palliative care for children with life-limiting conditions as "an active and total approach to care, from the point of diagnosis or recognition, embracing physical, emotional, social and spiritual elements through to death and beyond. It focuses on enhancement of quality of life for the child and support for the family and includes the management of distressing symptoms, provision of short breaks and care through death and bereavement"(2).

These definitions of palliative care for children are broad, and outline a philosophy of care (1, 2, 13). They imply that palliative care is a multidimensional, active process, which should occur alongside the management of a life-limiting or life-threatening condition. However, there is evidence to suggest that the term "palliative care" is inconsistently conceptualised and understood, which causes potential problems for patients, families and professionals. The term is often associated with a distinct time when cure-orientated treatment options have been exhausted (13-15). For some, it is inextricably linked to the end of life and dying. Inconsistent use and understanding of the term palliative care has implications in clinical practice, causing anxiety and acting as a barrier to conversations about the provision of palliative care. Referrals to specialist paediatric palliative care services often occur very late in the course of a child's illness, if at all (16-18).

Furthermore, there are guidelines and service specifications that refer to "palliative care" as a distinct specialist service (13, 19-21), creating further ambiguity about the definition of palliative care. There is longstanding debate about the elements of a healthcare service that comprise specialist paediatric palliative care. In England, the NICE Guidelines, published in 2016, define specialist paediatric palliative care services as those supported by a consultant with specialist training in paediatric palliative medicine (20), but many other models of specialist paediatric palliative care exist, including those provided by children's hospices, and nurse-led services.

The research conducted for this thesis aimed to examine the provision of palliative care as a broad approach to care that aims to improve the quality of life of children living with a life-limiting or life-threatening condition (1, 2), whether or not this involves the provision of care by a specialist paediatric palliative care team, or a children's hospice. Examining the delivery of palliative care in this way, rather than as a discrete intervention, raised particular methodological challenges. However, a clear aim of the research was to produce policy-relevant recommendations that have the potential to address current concerns about inequality in palliative care provision. Specialist paediatric palliative care services are not sufficiently developed or resourced to be the sole providers of palliative care to the rising number of children who could benefit from this approach. There is a need to "think outside the box" to consider the future design and delivery of palliative care for children.

Which children could benefit from palliative care?

Advances in medical treatments and technology are leading to a rapid rise in the number of children living with life-limiting and life-threatening conditions, both in the UK and internationally (7, 22, 23) 22). These children have a vast number of diverse diagnoses (24), often with long term, complex health and care needs (25). They live with uncertainty and a constant risk of a serious deterioration in health leading to an unexpected hospital admission, admission to the intensive care unit, or death.

Clinical tools to assist with the identification of children who have conditions where palliative care could be beneficial include a directory of life-limiting and life-threatening conditions (24), a categorisation of conditions (table 1.1) (2), and a spectrum of children's palliative care needs, which focusses on clinical signs that indicate a deterioration in the child's condition (26).

Table 1.1: Together for Short Lives Categories (2)

Category	Description
1 Life-threatening conditions for which curative treatment	Access to palliative care services may be necessary when treatment fails or during an acute crisis, irrespective of the duration of threat to life. On reaching long-term remission or following successful curative treatment

may be feasible but can fail	there is no longer a need for palliative care services. Examples: cancer, irreversible organ failures of heart, liver, kidney.
2 Conditions where premature death is inevitable	There may be long periods of intensive treatment aimed at prolonging life and allowing participation in normal activities. Examples: cystic fibrosis, Duchenne muscular dystrophy.
3 Progressive conditions without curative treatment options	Treatment is exclusively palliative and may commonly extend over many years. Examples: Batten disease, mucopolysaccharidoses.
4 Irreversible but non-progressive conditions causing severe disability, leading to susceptibility to impaired health.	Children can have complex health care needs, a high risk of an unpredictable life-threatening event or episode, health complications and an increased likelihood of premature death. Examples: severe cerebral palsy, multiple disabilities, such as following brain or spinal cord injury.

Who provides palliative care for children?

The thesis aims to examine specifically the contribution of healthcare services to palliative care for children. The provision of palliative care as a truly holistic approach extends well beyond the provision of healthcare services, to other statutory services including education and social care (19).

It is also important to consider that for children with life-limiting and life-threatening condition, parents or family members often become the primary caregivers. This can have a significant impact on their lives, health and wellbeing (27-29). Family members report that their role as expert caregivers for their child can be poorly acknowledged by healthcare professionals, and they draw on support from a wide range of support including friends and peers, both online and in person (30).

Paediatric palliative care: a developing specialty

The provision of specialist paediatric palliative care services to children and families is inconsistent and inequitable in the UK and internationally (23, 31, 32). Furthermore, there is emerging evidence that children with life-limiting and life-threatening conditions and their families have a wish for continuous, holistic healthcare, with an option for that care to be delivered in the home environment, but this is often difficult to achieve (33).

The medical specialty of palliative care has developed over the past 50 years, pioneered by clinicians who witnessed distressing deficiencies in the care of patients at the end of life. St Christopher's Hospice in London, the first organisation built specifically for the care of dying adults, opened under the direction of Dame Cicely Saunders in 1967. The approach to care championed by Saunders was holistic person and family care, delivered through multi-disciplinary teamwork. Balfour Mount, a surgeon, introduced the term "palliative care" in 1973 as he created a palliative care ward for the care of the dying at the Royal Victoria Hospital in Montreal, Canada. Much of the palliative care

provided in these specialist organisations was for adults with cancer, who required pain and symptom management at the end of life (34). Over the last 20 years, as clinical need has shifted and changed, the palliative care needs of adults with long-term incurable conditions such as heart failure, chronic obstructive pulmonary disease (COPD), dementia and multi-morbidity have been increasingly recognised (14, 35, 36).

Palliative care for children has grown alongside developments in adult palliative care, through the opening of children's hospices and the recognition of paediatric palliative medicine as a sub-speciality of paediatrics. Many paediatric palliative care services in the UK, including children's hospices, have not developed through any national strategic approach, but because of the determination and drive of individuals, with sporadic funding opportunities often provided by the voluntary sector (37). Many children's hospices originally existed to provide respite care; however, there is now an increasing need for expertise in complex symptom management for children (13, 38, 39). Paediatric palliative medicine first became a sub-specialty of paediatrics in the USA and Canada (40). It has been a sub-specialty of paediatric medicine in the UK, recognised by the Royal College of Paediatrics and Child Health (RCPCH) and with a specialist training programme, since 2009 (13).

Despite this progress, the provision of effective, equitable palliative care for all who need it remains a pressing concern. A series of high profile events over the past five years (through the course of this PhD) have highlighted continued deficiencies in the care of the dying. These have included significant concerns about the use of the Liverpool Care Pathway (41), and the publication of a report by the Parliamentary Ombudsman, "Dying without Dignity" (42), both of which have attracted significant media attention and resulted in a plethora of new policy documents. Although the focus has been on adult palliative and end of life care, many of these policy documents are relevant to the provision of palliative care for children. The national strategy for England, Ambitions for Palliative and End of Life care, places specific attention on the need to improve palliative care for children (43).

Commissioning palliative care for children
Current guidelines and models of palliative care for children describe the need for the commissioning of services across three levels of palliative care (19). This is a widely accepted model, described in children's palliative care for several years (38) (outlined in Figure 1.1).

Figure 1.1: Three levels of children's palliative care

**Specialist
Palliative Care**
(hospital, home or
community)

Core Palliative Care Services
These form the majority of services
required by children and young people
with palliative care needs (e.g. local
hospital, community paediatrics,
community children's nursing teams,
children's hospices, children's palliative
care charities)

Universal Services
The foundations for good palliative care include health and
social care services which are available to all children and
young people (e.g. public health, GPs, education, social
workers, playgroups and the wider community)

At the top of the pyramid are specialist paediatric palliative care services. The base of the pyramid comprises "universal" services, described as "the foundations for palliative care". In the middle of the pyramid are "core" palliative care services, those providing the majority of services and care for children with palliative care needs, including children's community nursing teams and paediatricians. The voluntary sector, children's charities and hospices, are included as core palliative care services. In England, commissioning specialist services, including specialised paediatric palliative medicine, is within the remit of regional NHS England Specialised Commissioning teams, with responsibility for universal and core services lying with local Clinical Commissioning Groups (CCGs) (31).

Why should we improve palliative care for children?
Clinical, ethical and political imperatives exist for improving the delivery of palliative care to children, which require urgent attention as the numbers of children living with life-limiting and life-threatening conditions rises. Difficult clinical and ethical issues arise due to the impact of the condition on the child and their family, and balancing the quality of life of the child with the delivery of intensive, experimental or invasive medical treatments and technology. Ensuring that the healthcare they receive meets their needs, and makes best possible use of healthcare resources are also pressing concerns.

The numbers of children who die in the UK remain relatively small when compared to the adult population (44). However, over half of children who die have a pre-existing life-limiting or life-threatening condition (45), most die in hospital (46), and many die in an intensive care environment, where the mode of death is often withdrawal of life-sustaining treatments (47-49). The length of stay in the intensive care unit before death is increasing, reflecting a trend towards longer attempts to sustain life (50). Weighing up the benefits and potential harm of prolonged intensive care towards the end of a child's life is clinically and ethically challenging, and can cause significant distress for both staff and family members (51-53). Several high profile cases have attracted intense media attention during the course of this PhD, perhaps most notably the cases of Charlie Gard, Alfie Evans and Isaiah Haastrup (54-56). These cases illustrate the intensely complex influences that now affect the provision of healthcare for children with life-limiting and life-threatening conditions who may die. The quality of life of the child and their family, the availability of new and experimental treatments, funding for healthcare costs, public perceptions and the influence of both the media and social media all influence decision making.

1.4. Motivation for research

Like many other clinicians, my interest in palliative care stems from witnessing deficiencies in the provision of healthcare to people with life-limiting and life-threatening conditions, and those who are dying, in my clinical practice.

I encountered a dying child for the first time in my career on a cold, dark and rainy night while working as a junior doctor in paediatrics as part of my general practice vocational training in 2004. I was one of a team of doctors and nurses in the emergency department when a seven-year-old boy, with a severe neurological condition, arrived in the department with his mother. He was critically unwell with signs of a lower respiratory tract infection, a condition with which he had been into hospital several times over the preceding weeks. We took him straight through to the resuscitation bay, and crowded around him, giving him oxygen, taking blood, and inserting cannulas and an arterial line. I will never forget his mother in the corner of the room, asking us all to stop. None of us did, until his usual consultant arrived in the room. Only then, did we stand back and stop trying to deliver painful and invasive procedures. The child was admitted to a medical ward, and he died there, with his family around him, two weeks later.

I have now been qualified as a general practitioner (GP) for 12 years, and have encountered a number of other memorable children with life-limiting and life-threatening conditions. In 2010, when I was working as a GP partner, a child who was a patient at the practice died. She died in the regional paediatric intensive care unit (PICU) following months of repeated severe deteriorations

and admissions to PICU, and I was the GP to receive the letter notifying us of her death. On review of her medical notes, I discovered that she had been living with two long-term life-limiting conditions, both diagnosed during her early childhood. I remember feeling appalled that as her GP practice team, we had not managed to recognise the nature of her conditions, included her on our palliative care register or provided any proactive support to her family.

My motivation to undertake research and work to improve children's palliative care is grounded in this clinical experience. In my work as a GP, I have witnessed not only a rise in the complexity of the patient population, but also changes in the healthcare service which have fundamentally altered the way in which we deliver care. Changes in primary care contracting and the organisation of community care services alongside the specialisation of hospital medicine have resulted in fragmented services with fewer opportunities for the provision of proactive holistic care, including palliative care. This is despite the plethora of policy guidance calling for improvements in palliative care provision. With general practice being under "unprecedented pressure", and strict referral criteria for specialist services that do not always meet the individual needs of patients, there are many barriers to the delivery of proactive holistic care for those with the most complex needs. I have been motivated to undertake this research by a recognition of the increasing medical, social and psychological complexity of the population, and a need to consider different ways in which to deliver healthcare so that the palliative care needs of those, particularly children, with life-limiting and life-threatening conditions are met, and their experience of healthcare improved.

1.5. Rationale for research and initial programme theory

The number of children with life-limiting and life-threatening conditions is rising. The complexity of their medical conditions is increasing, as are their associated care needs. Specialist paediatric palliative care services are patchy and inconsistently resourced. Despite the range of policy recommendations for the provision of palliative care to children and families, there remains a lack of research evidence to support the implementation of policy and guidelines in practice.

The rationale for this research is to contribute to the evidence base to inform future service design and policy in palliative care for children. The research holds the children and their families at the centre, and focusses on their experiences of the delivery of healthcare, and in particular palliative care. The research starts with a theory, drawn from the background provided, through review of national and international policy documents, and from my observations in clinical practice, that palliative care for children "works" to improve their quality of life and that of their families.

1.6. Aim of the research

The aim of the research is to provide new insights and understanding into the healthcare experiences of children with life-limiting and life-threatening conditions, and their families, in order to contribute to the evidence base and consider new approaches to the provision of palliative care to children in the future.

The research has been designed to test the initial programme theory that palliative care "works," generate new theories and lead to the development of tangible, policy-relevant recommendations for the future delivery of palliative care for children. This has involved the following stages:

1. Development of research questions, informed by patient and public involvement, that correlate with current concerns associated with the delivery of palliative care to children and their families.
2. Systematic appraisal of existing policy, guidance and published evidence related to palliative care service delivery for children with life-limiting and life-threatening conditions and their families.
3. A qualitative investigation into the healthcare experiences and preferences of key stakeholders, including the perceived facilitators and barriers to the delivery of palliative care. Key stakeholders included children with life-limiting and life-threatening conditions, their family members and paediatric palliative care professionals.
4. Thematic analysis of the findings, followed by application of a realist logic to this analysis.
5. Generation of context-mechanism-outcome configurations (CMOCs) and an overarching programme theory to inform policy-relevant recommendations for the future delivery of palliative care for children.
6. Patient and public involvement work to inform the research throughout.

Figure 1.2: Summary of plan for research
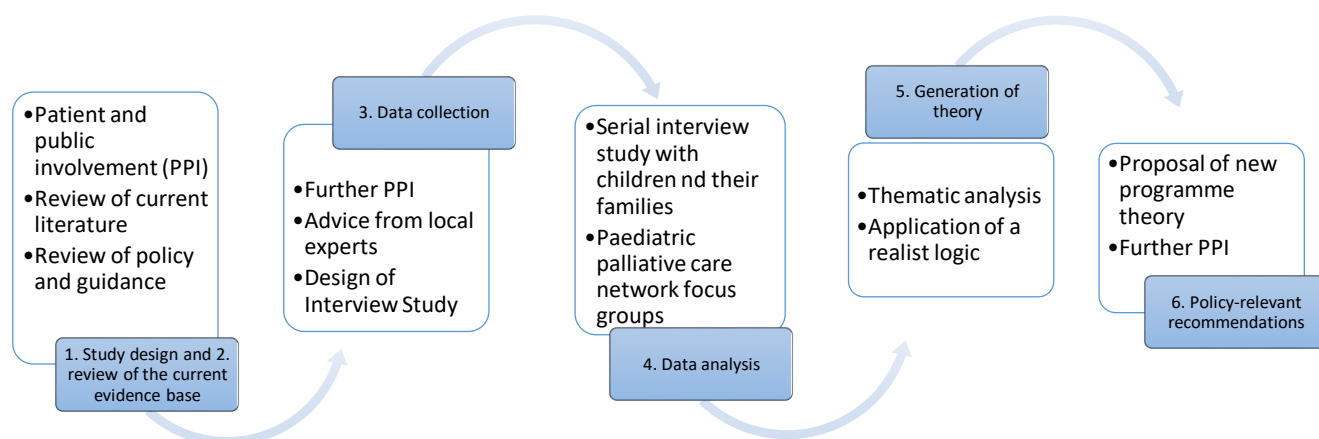
1.7. The research questions

The research aims to address the following questions:

1. How do current definitions of "palliative care" for children concord with service delivery, policy and guidance in the UK?

2. What is the current evidence base for practice and policy related to palliative care service delivery for children?

3. What are the lived experiences of children with life-limiting and life-threatening conditions and their families?

4. How do children with life-limiting and life-threatening conditions and their family members perceive healthcare services, including "palliative care"?

5. When and how does "palliative care" provide benefit for children with life-limiting and life-threatening conditions and their families?

   1. What outcomes are important to children and their families?

   2. What are the mechanisms by which these desired outcomes are achieved?

   3. What are the contexts that determine whether or not these mechanisms produce the intended outcomes?

1.8. The study title

Young people at Acorns Children's Hospice chose the study title, "The Journey through Care", during a patient and public involvement (PPI) session early on in the course of the research. All of the young people involved in the session lived with a life-limiting condition and had received hospice services. They rejected the original title for the research, "Palliative Care for Children and Young People: What? When? How?" expressing concerns about the term "palliative care". They were not at all

familiar with the term, did not understand it and did not view it positively or want to relate it to themselves or their friends. Instead, they felt the study should reflect the reality of their "journey" through their condition and interactions with the healthcare system, which was associated with significant "ups" and "downs", was highly unpredictable, and had an uncertain ending.

1.9. Chapter Summary

This chapter has provided a description of the current situation in the provision of palliative care to children and described why there is a need for more research in children's palliative care. The rationale for the research has been explained, along with the aim of the research, to test an initial programme theory that palliative care "works". My own clinical experiences in the care of children with life-limiting and life-threatening conditions, and their families, have been some of the most formative of my career, and provided the motivation to undertake this study. PPI has been an important and integral component of the research throughout, and the study title reflects the impact of this PPI work. The PPI is described in Chapter 2.

2. Patient and Public Involvement

2.1. Overview of Chapter 2

This chapter describes the Patient and Public Involvement (PPI) work that was integral to the study. The GRIPP2 reporting checklist, which outlines the key items to report to enhance the quality, transparency and consistency of the PPI evidence base has been used to provide a structure for the chapter (57). Throughout this chapter, the terms "children and young people" or "young people" have been used. This seems more appropriate than referring to those who took part in the PPI work, who were aged between nine and 25 years, as "children".

2.2. Definition of Patient and Public Involvement

For the purposes of this study, the INVOLVE definition of PPI was adopted: the active involvement of patients and members of the public in the design and process of research to ensure that it is relevant and contextual (58). This is different to participation in research and public engagement activities related to research.

2.3. Background

PPI is recommended at all stages of the research process, from creating the initial research questions to specific aspects of study design, data analysis and dissemination, and has been shown to have a positive impact on research, with studies with PPI more likely to recruit to target (59, 60).

The active involvement of patients and the public is also gaining prominence in service design and commissioning in healthcare. The aim of this involvement is to ensure that the people who use services and their experiences are at the centre of decisions about the design of future care services, frequently through a process of engagement or public consultation. Despite the increasing policy agenda and prominence of PPI, this remains a developing field with a variety of different approaches taken, many of which have been criticised for being exclusive and tokenistic (61). PPI can be challenging for researchers, with no agreed best practice approach and the possibility that it will raise unanticipated issues.

The importance of the active involvement of children and young people in research that concerns their care is increasingly recognised (62-64), and is a specific focus of INVOLVE and the work of the Royal College of Paediatrics and Child Health (RCPCH) (65-67). Children and young people can make valuable and highly relevant contributions including insights into a child's worldview, contemporary understanding of influences such as social media, and input that ensures that the research design is appropriate for participants who may be at a similar developmental stage to PPI group members. There are a number of benefits for those who are involved including a feeling of empowerment,

increased confidence and self-esteem, gaining new knowledge, skills and experience, and working with peers (68).

The reporting of PPI in research has been inconsistent to date. Sharing of information about theorisation, context and impact of the PPI process has been limited. As a result, there have been limited opportunities for learning from the experience of others or any move towards a consensus for the most effective ways to conduct PPI (57). There is a range of guidance to support the conduct of PPI, including specific resources to support PPI with children and young people (65, 69). There are also tools to assess the impact of PPI, however the evaluation of these to date is limited (65, 70-73). Currently the evidence base to inform the conduct of PPI with children and young people is limited (74, 75), with much of the published literature focussing on the experiences of children and young people as research participants (76-80) rather than their experiences of PPI work or the impact that this has on the research.

Aims

The aims of PPI for this study were:

1. To collaboratively involve children and young people with a range of backgrounds and experience at all stages of the research, from study design to dissemination.
2. To develop an ethical approach for PPI with children and young people.
3. To contribute to the evidence base for PPI with children and young people, and for palliative care research.

2.4. Experiences and impact of PPI

PPI was integral to the study design and had demonstrable impact in several areas. The initial research idea and the plan for research, including the practicalities of conducting interviews with children was informed by PPI. PPI representatives played a part in developing the recommendations from the research and took part in a range of dissemination activities. PPI also led to new ideas for research, including an idea for a survey study about the language of palliative care, described in more detail below.

PPI that informed the research plan and procedures:

The initial research idea and funding application were both informed by a PPI workshop that was held in March 2013. Three parents of children with life-limiting and life-threatening conditions, and one young person attended. The workshop was held at Acorns Children's Hospice, and was facilitated by one of the PhD supervisors (JC). The views and ideas of those who attended were captured using flipcharts and postcards. These were subsequently collated, and used to inform the

development of the research questions and the application for the Doctoral Research Fellowship (DRF).

Group members provided advice on the practicalities of the study design including the design and wording of participant information leaflets. They advised on the conduct of interviews with children and on the wording of questions in the topic guide. They designed a logo for the study and the posters used for recruitment.

Development of recommendations:
A presentation of the research findings was given to the NIHR CRN Young Person's Steering Group (YPSG) in January 2019. The children and young people at the meeting provided reflections and written feedback on what they considered to be the most striking and important points. These were used in the development of the recommendations from the research, and will be used to inform a dissemination film.

Dissemination:
PPI group members were also involved in the design of conference posters, and took part in presentations at regional and national conferences, summarised in table 2.1. They also ran a workshop, "Involving children in research about sensitive subjects" at the RCPCH conference in Glasgow in 2018. Three of the group members co-authored journal articles, suggesting edits and reviewing the content to ensure that the articles were relevant to the child and family experience.

Table 2.1 Summary of dissemination activities with PPI group members

Year	Conference and presentation	Type of presentation	Young people involved
2016	RCGP Mid Faculty	Oral	Laiba and Sophie
2017	RCPCH Annual Conference (National)	Poster	Mohini, Olivia and Clare
2017	SAPC	Oral	Mohini
2017	ICPCN	Oral	Mohini, Olivia, Clare, Dena, Zack
2018	RCPCH	Workshop	Mohini, Dena and Lizzie
2018	NHSE event	Stakeholder event	Molly
2019	NIHR Hospice Consortium conference	Oral	Sophie

Photograph 1: A PPI session
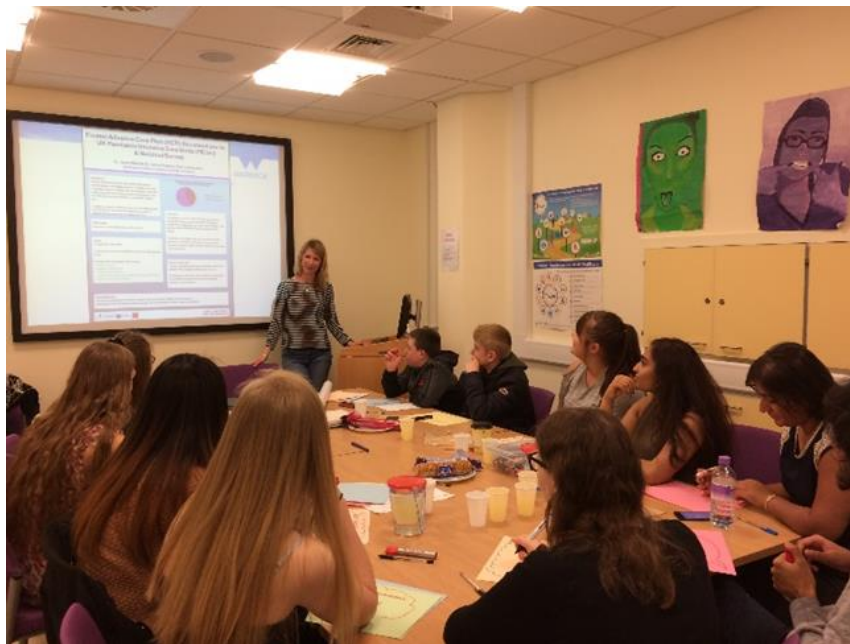
Photograph 2: PPI group members with their poster at the 2017 RCPCH National Conference, Birmingham

Photograph 3: PPI group presentation at the 2017 International Children's Palliative Care Network Conference, Cardiff

Photograph 4: RCPCH 2018 conference workshop preparation

Further research and working as co-researchers:
The PPI work generated new research ideas, particularly related to the understanding of the term "palliative care" amongst children, young people and healthcare professionals. Together, we devised a research protocol for a survey study of children, young people and healthcare professionals from

five children's hospitals in the UK. One of the young people attended the research ethics committee (REC) meeting (Dena, who has written a reflection below). Another young person, Dan, helped with data collection at his school, and another, Sophie, arranged summer work experience as a Research Assistant at the University of Warwick. In doing so, she was able to use the skills in qualitative data analysis that she had learnt during her first year at university to carry out thematic analysis on the children's survey results, and work on drafting an academic paper of these findings for publication.

A problem we encountered with the conduct of the survey study was in the recruitment of young people from a secondary school. By the time ethical approval was granted, the young person who had volunteered to take the survey in to her school had gone to university. The Research Ethics Committee (REC) advised a need for a major amendment to the protocol in order to recruit participants from another school. Given the scarce time resource of the research team (myself and the young people), we agreed not to proceed with this, but instead analyse the results of the survey responses from the primary school and two other groups of young people.

Ethical considerations
In the UK, there is no requirement for ethical approval when undertaking PPI for research. Ethical guidance relating PPI with children and young people is scarce. Given the potentially sensitive subject area of this research, an ethical approach to the PPI work was devised, followed and has also been published (Appendix 1 (3)). Table 2.2 provides the key principles of this ethical approach to PPI:

Table 2.2: An ethical approach to PPI: key principles

Step	Ethical approach
1	Prioritise PPI with children and young people
2	Agree language and work towards a shared understanding of tasks
3	Gain consent for PPI
4	Maximise the benefits for PPI group members
5	Minimise the risk of harm
6	Ensure equity of access to PPI
7	Provide training for the researcher
8	Offer training for the PPI group
9	Provide funding and recognition

2.5. Methods

Participants

PPI was carried out with members of existing groups of children and young people; Birmingham Children's Hospital (BCH) Young Person's Advisory Group (YPAG), the NIHR Clinical Research Network (CRN) West Midlands Young Person's Steering Group (YPSG) and Acorns Children's Hospice, West Midlands. Members of these groups ranged in age from nine to 25 years. They had a wide range of personal experience and included young people who currently lived with a life-limiting condition or had previously received treatment for a life-threatening health condition such as cancer. Others lived with a seriously unwell sibling or were bereaved of a sibling or cousin. The young people had volunteered to be members of the groups because they had particular interests in research, service improvement or policy.

Procedures

PPI was an integral element of the research from the early stages. Table 2.3 provides detail of the PPI activities that were carried out at each stage of the research:

Table 2.3: The PPI timeline

Date	Activity	Supported by	Impact
Mar 2013	Workshop with three parents and a young person. Highlighted the vast number of priorities for research from the family perspective, particularly health systems research.	Acorns Children's Hospice & Prof Jane Coad	Views informed the aim of the research and research proposal. PPI section of application form completed.
Nov 2014	Meeting with two young people with life-limiting conditions: challenged the term "palliative care" and the study title.	Acorns Children's Hospice	Title changed to "The Journey through Care" Idea for survey of young people and development of further research questions
	Meeting with the sibling council. Eight young people. Introduction to the study with leaflets to take away, conversation about research and to establish level of interest in this research.	Acorns Children's Hospice	Ongoing challenges with language and need for succinct summary of research in plain English (no jargon). Realised the value of capturing written as well as verbal feedback.
Jan 2016	Meeting with 22 young people: checked understanding of the study, general feedback given on the study design and specific details related to the design of	BCH YPAG	Informed design of participant information sheets for younger and older children

	participant information sheets. Written and verbal feedback provided.		
May 2016	RCGP Midland Faculty conference presentation: 2 young people attended and co-presented	BCH YPAG and Acorns Children's Hospice	Presentation on ethical approach to PPI given with young people
Jul 2016	Meeting with 8 young people. Logistics of interviews discussed.	NIHR CRN	New interview questions devised and tested.
Oct 2016	Idea for language survey discussed and questions for questionnaire considered	Acorns Children's Hospice	PPI research project devised (survey about language of palliative care), protocol written.
Jan 2016 – Jan 2017	Young person volunteered to take part in writing a systematic review, providing family perspective.	Acorns Children's Hospice	
Feb 2017	Further discussion about project design and update on progress with interviews. Discussion of dissemination strategy.	BCH YPAG	YPAG have close links with hospital managers, to be considered in dissemination activities.
April 2017	PPI session with 14 young people: poster design discussed	NIHR CRN YPSG	Poster designed for RCPCH conference
May 2017	RCPCH conference: poster presentation (Birmingham International Conference Centre).	NIHR CRN YPSG	Three young people attended the RCPCH conference to present the poster
Jul 2017	Society of Academic Primary Care conference presentation (University of Warwick)	NIHR CRN YPSG	One young person and I presented on young people on their experiences of PPI for this palliative care research
Jul 2017	International Children's Palliative Care Network conference presentation (Cardiff University)	NIHR CRN YPSG	Five young people took part in the presentation on their experiences of PPI for this palliative care research
Aug 2017	East Midlands REC meeting re. survey proposal	NIHR CRN YPSG	One young person (Dena) attended the REC meeting

Nov 2017 – Mar 2018	Survey project carried out. Recruitment of children and young people from Acorns, a local primary school and BCH YPAG.	NIHR CRN / Acorns Children's Hospice / BCH YPAG	Research designed with young people from the PPI groups. Young people recruited as participants.
Mar 2018	"Should we involve children and young people in research on difficult topics: a chance to ask them". Workshop delivered with three PPI co-presenters at RCPCH conference (Glasgow).	NIHR CRN	Three young people presented a workshop at the conference
Nov 2018	Attendance at NHS England national commissioning model for children's palliative care stakeholder event as the only young person representative	NIHR CRN YPSG	One young person attended a national commissioning meeting
Jan 2019	Research findings presented to the group with feedback sought on the presentation, developing recommendations and ideas for a dissemination film	NIHR CRN YPSG	Discussion with the group about the findings of the research, and how best to present these. Early plans for a dissemination film discussed.
Next steps	Dissemination and film to be produced	NIHR CRN YPSG	2019/20
	PPI for future research proposals	All	Extend PPI opportunities to other young people beyond these groups.

During the course of the study, PPI advice was actively sought at regular intervals by attending existing group meetings, and running task-orientated sessions with activities that had been carefully designed at each stage to capture the contributions of the group.

The structure of sessions developed iteratively as I learnt what worked and what didn't work to gain useful feedback and input from the group sessions. Ensuring a clear aim, with a pre-prepared, specific, structured task for each session was effective. A flexible approach was necessary in order to fit in with each meeting agenda, the expectations of group facilitators, and to respond to new ideas raised by the young people during the sessions. Notes were taken during and after each session and written feedback forms were provided to the group so that further contributions and reflections could be captured. This was particularly important to ensure that the views of quieter group members were captured. Further PPI was conducted between meetings with specific points clarified via email and documents or presentations devised using shared documents on GoogleDocs.

2.6. Evaluation

Little is known about the experience of children in PPI for research. An evaluation of their experiences of PPI for this research was conducted using a method based on the "Tell Me…" exercise outlined in RCPCH &Us Recipes for Engagement. This involved group members providing anonymous feedback about any aspect of their experience related to the PPI for this research on post-it notes (69). There was no restriction in terms of word count in their responses, and they were invited to use as many post-it notes as they wished to. All comments were anonymous with no information requested that would identify the individual who had written the comment. The feedback was transcribed and imported to NVivo data management software. An inductive thematic analysis was carried out, assigning every piece of feedback to a category, then grouping these categories into themes (81).

Findings of the evaluation

30 young people provided comments during three meetings held between December 2016 and August 2017. The youngest participant in the evaluation was 12 years of age; the oldest was aged 22 years. Six of the young people had previous personal experience of palliative care (four from Acorns Children's Hospice, and two from the NIHR CRN group). Three key themes emerged from the data: firstly, the young people wanted to be involved, secondly, they wished to have impact on the research, and for the research to have impact, and thirdly, they were keen to learn from their involvement, described in more detail below as (1) involvement, (2) impact and (3) learning.

1. Involvement

Young people expressed a desire to be involved in the research despite the potentially sensitive subject area of palliative care. No group members opted to stay out of presentations or leave group sessions. Motivation to be involved included the opportunity to voice an opinion, and to make a difference: *"It's amazing being involved, allows us to voice our own opinions and to be given the chance to make a difference".* Others expressed a desire to wanting to help others, and the community: *"Helps to make you feel that you are involved in helping the community".*

Several young people expressed a desire not to be excluded from conversations about palliative care, recognising the importance of the subject area. One young person stated: *"Really exciting! Important: so often overlooked or side-lined or delayed referral as treatment is often seen as superior to palliative care".* They acknowledged the presence of societal barriers to discussing palliative care openly: *"Important as no-one wants to talk about it!"*

One young person expressed very personal reasons to contribute to the PPI, explaining that she often felt excluded from healthcare decisions for her sister, who had a life-limiting

condition. She stated, *"Being asked about palliative care is very interesting because as a young person I am interested in what happens to my sister and the choices that are made and I am not normally involved when I would like to be. This research project gets my own opinions and thoughts about palliative care which is good as it means I am involved and listened to for once."*

2. Impact

Young people wanted to see that their input had impact on the research plans, and that the research had the potential for impact on policy and practice. They expected to be listened to, and wanted to hear from me how their advice and views had influenced the research process. There was clear instruction to return to the group and inform them of progress: *"Ensure we receive feedback and follow through throughout the project."*

There was value in the development of an ongoing relationship between myself as the researcher, and the PPI groups throughout the research. This allowed rapport to develop, and an open, conversational approach to the group sessions that allowed the discussion of sensitive topics, as illustrated by the following quote: *"it* [the PPI] *has been conducted in a way that makes me comfortable to contribute."*

3. Learning

Young people described benefits of taking part in PPI for this research as opportunities to learn about the topic: "*I think this project is very interesting and I can't wait to hear more about this. I don't know much about palliative care so I'm keen to learn more about it."* They viewed involvement in research as a way of learning, which had the added benefit of helping someone: *"It's good to do a research project because it gives you knowledge of the subject and you know you're helping someone or something."*

Young people's accounts of their involvement

This section of the chapter comprises a series of reflective paragraphs, written by members of the PPI group who were involved in certain aspects of the study. The paragraphs represent a range of experiences, both positive and negative, related to aspects of the PPI from the perspectives of the young people who took part.

Box 2.1: Acting as a Co-Author on a paper, Laiba Sajid, Acorns Sibling Council

> When I first read the paper I felt really overwhelmed and honoured that I was able to be part of this. I felt that my voice was important and I was speaking for many in similar circumstances to me. What I felt was that I can be part of something very important and my voice will be heard.

I would say to other young people who are asked to co-author a paper, do not be scared and say how you feel and be honest because if we are not asked or do not say what we feel then how are things going to change? It is being the voice of many.

I felt really honoured that I was even asked. For me it is being part of something that could make a difference. I will never forget this experience and it has also given me an insight into how things can be changed for the better.

Box 2.2: Attending a conference, Clare Atherton, NIHR CRN YPSG

My first conference was the RCPCH annual conference and it was exhilarating. I had only been a part of the Young Persons´ Steering Group for a few months. Olivia (also from the group) and I were presenting a poster about the importance of PPI. Initially I was a little bit uncertain but I wanted people to think I was approachable. The first question I was asked completely stumped me and I felt like I had no idea what I was doing! Even so I was soon assured by the positive feedback we were getting and felt much more comfortable talking about the poster.

I was really honoured to be given this opportunity as I hope to train to be a doctor in the near future and I found it really interesting to network with everyone there. It made me realise that there is much more to research than the stuff we see as a group. Being able to share my passions around the Young Persons´ Steering Group and learn new things has been amazing.

One of the best things about the conferences I have been to is that I have been regarded as highly as all the professionals there. I have felt as though my voice has not just been heard but people have listened actively. I felt as though everyone at the conference was really engaged with me and the work I have been talking about. It is definitely an experience I hope to repeat many more times.

Box 2.3: Attending a research ethics committee, Dena Khan, NIHR CRN YPSG

The prospect of partaking in an ethics committee was an exciting opportunity. My understanding of clinical research has allowed me to understand the importance of ensuring any form of research is ethically sound. I want to pursue psychology and psychological research so this experience was even more valuable to me.

Having no clue what to expect, I found the event insightful and interesting, although it didn't take very long! I was able to see how important the ethics approvals process is to hold researchers to account and make sure patients/participants are remaining the central focus of any study.

I was reassured that our study did not prompt a lot of ethical concerns, and I felt glad to be part of a project that takes into account both our opinion and the welfare of those involved.

Being able to go to an ethics committee has furthered my interest in research, and has made me grateful for the amount of precautions put in place. However, it has also shown me how young people can be so easily involved in research and how our opinions and ideas can be used to the benefit a study as I noticed the surprise in the committee of a young person's presence.

Box 2.4: Attending an NHS England Commissioning Meeting, Molly Seaborn, NIHR CRN YPSG

My experience overall summed up in one word would be enlightening, in two ways. The first was that I learnt so much about the way commissioning models for healthcare are created. It gave me an insight into the world of NHS England and I felt honoured to be (a small) part of such important work for young people's palliative care. However, the second way was that I was exposed to a form of discrimination that I had not expected since I had been invited as a voice for young people. As the only young person in a room of professionals, I was disappointed that it seemed my opinion was discredited without thought by some, despite what I believed to be valuable insight that they had no other means of access to. It felt as if my age was grounds for being ignored and even mocked at times. However my experience overall was positive as I feel I may have been able to change even a couple of people's minds about young people being involved in the commissioning of our care services and because it was an excellent and rare opportunity to hear about an area that I am interested in.

Box 2.5: Inspiring PPI group members, Mohini Samani, NIHR CRN YPSG.

I am currently in the process of picking my dissertation topic … and due to the work I have done with you I have decided to explore the historical aspects of palliative care in NHS England. I was planning on emailing you soon to ask that as part of my dissertation I have to do primary data collection in the form of interviews, so if I could interview you and if you could put me in touch with other people who work in the field. This is looking quite far into the future but thought I would ask in advance.

2.7. Discussion

There are very few papers documenting the experience of young people in PPI for research. One of the aims of the PPI for this research was that children and young people from a range of backgrounds and experience would be involved in the research at all stages, from study design to dissemination, and that their autonomous views would be valued and heard. This was achieved by approaching existing groups of young people at different stages of the project, who were meeting at a time that was convenient for them, to gather their views and opinions as the research progressed.

PPI input has enhanced aspects of the study design and the conduct of the research. PPI group members provided constructive criticism and new ideas that challenged my own. They provided helpful advice and valuable insights that contributed to developing the research plan and resources such as participant information sheets. There was particular enthusiasm for taking part in dissemination activities, with eight young people taking part in oral presentations at national and international conferences, and two more presenting at regional conferences.

Strengths, limitations and learning
The key characteristics of effective PPI within individual research studies have been described in previous research as (82):

- A shared understanding of the moral and methodological purposes of PPI
- A key individual co-ordinating PPI
- Ensuring diversity (of PPI representation and inputs)
- A research team positive about PPI input and fully engaged with it
- Relationships that were established and maintained over time, and
- PPI being evaluated by a proactive and systematic approach.

The moral and methodological purposes of the PPI for this study were set out at the early stages. There was a commitment by the research team to ensure that PPI was integral to the conduct of the study, that the approach would value the autonomous contributions of young people and that it was conducted in an ethically sound manner. The young people who were involved had a diverse range of life experiences, views and opinions, and the PPI was carried out at existing meetings, in order to fit in with their commitments. As the PhD researcher, I led and co-ordinated the PPI activities, and established relationships with the PPI group members that were maintained over time.

Accessing existing groups also had the advantage that they were supported by a group facilitator. However, access to the groups depended on engaging these group facilitators, and ensuring a shared understanding of the purpose of the PPI. This was more difficult for those who were not familiar with research. Engagement was most successful with the NIHR CRN young person's steering group, a group which exists with the purpose of involving young people in research. For the other groups, where the purpose was not specifically involvement in research, this was more challenging, and depended on the perception of the group facilitator of the importance of research. For one of the groups, there was a regular change of group facilitator. With each change there was a change in the structure of the group meetings and a need for me as a researcher to engage the new facilitator. The enthusiasm of the facilitator and willingness to discuss research with the group depended on these individuals. An important consideration of approaching existing groups was the issue of equity of

access to opportunities to become involved, which are not widespread currently (68, 83, 84). The groups were dynamic with changing membership over the course of the study, suggesting that there was success in recruiting new members. A further advantage of accessing existing groups is that it was cost effective. Since the group meetings were already established, the PhD PPI budget was used to enable other PPI activities, for example paying for conference fees and travel to meetings and conferences.

Approaching different groups of young people for PPI was conducive to gaining a wide range of views and opinion but is different from conventional approaches to PPI, where a project-specific PPI group is recruited. Ensuring an approach to PPI that fits with the busy lives of young people is important. As well as school or university commitments, some young people balance their involvement with management of their own health concerns and hospital appointments. For tasks that required a small group, for example the presentation at the International Children's Palliative Care Network (ICPCN) conference, we used online methods for conversation and development of ideas for the presentation, rather than meeting face-to-face. Taking this approach allowed the young people to take part at times that were convenient to them.

A specific aim of the PPI was to develop an ethical approach to PPI with children and young people. This was particularly relevant given the potentially sensitive subject area of the research. The ethical approach we proposed and published is widely applicable across other research and service improvement projects.

At one of the early PPI meetings, group members requested that a Twitter account for the study was requested to keep them informed of the study progress. The account was created, but its use was limited in terms of both posting updates and interaction over social media with group members. Use of social media for PPI with young people warrants further attention and exploration.

Evaluation of PPI is not widespread. We adapted a method which had been devised and tested by young people and allowed for detailed anonymous feedback to be provided on all aspects of the PPI work (69). The qualitative data provides insights into how young people experience PPI, and their experiences related specifically to palliative care research. Young people expressed a desire to be involved in the research despite the sensitive subject area. Both the relationship with the researcher and the environment in which the PPI was conducted were important in order for them to be able to express their views.

Researchers can be criticised if they fail to engage or update young people as the research progresses. There is a need to be mindful of group expectations, PPI group members may be more

optimistic about the impact of their input than the researcher (85). Terms of reference or an agreement drawn up between the researcher and PPI colleagues early in the process would be a proactive approach to this. In keeping with other studies (86, 87) the young people who took part in the PPI evaluation expressed a desire to be heard and for the PPI to not be tokenistic. They wanted to be listened to and to see that their input had impact. Returning to the groups to update them on the progress of the project, involve them at each stage and to inform them of how their input had affected the research plans and conduct was important. This research study was Highly Commended at the NIHR CRN West Midlands Awards in 2017.

2.8. Chapter Summary and Conclusions

The PPI for this research was a valuable and fulfilling experience. The relationship with the researcher and the conduct of the PPI work were both important factors. The young people were motivated by a desire to learn and influence the research process, as well as for the research to have impact. There are particular challenges and opportunities in the conduct of PPI with young people including conducting PPI in a way that is convenient for them, and ensuring the approach to PPI is ethically sound. There is an ongoing need to share best practice and evaluation of PPI in research, to ensure that approaches are robust and meaningful, for wider opportunities for involvement and for a higher representation of PPI in research dissemination.

3. Literature Reviews

3.1. Overview of Chapter 3

Two systematic literature reviews are presented together in this chapter. The first was a systematic literature review (published in 2017 (Appendix 1 (4)). This review examined evidence relevant to the current policy recommendation for specialist paediatric palliative care services, defined as those supported by a consultant with specialist training. The second review was a realist review, the aim of which was to investigate "when" and "how" palliative care provides benefit to children with life-limiting and life-threatening conditions and their families. A protocol for the realist review has been published in PROSPERO (registration no: CRD42018090646 https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=90646), and the review published in August 2019 (Appendix 1 (5)).

The research teams for both of the reviews comprised myself, the supervisory team (JD, A-MS, and JC), and two medical students, Karina Bennett (KB) and Andrew Morris (AM). Karina and Andrew undertook elements of the reviews for their selected student component projects. Both of the reviews have been updated for presentation in this chapter.

3.2. Patient and Public Involvement

Both of the literature reviews began with ideas generated through the Patient and Public Involvement (PPI) work outlined in Chapter 2. The research questions for the reviews were informed by the views of PPI group members, volunteered during group meetings at the start of the PhD. One young person, Laiba (LS), from Acorns Sibling Council became more involved in the systematic review in 2015, when she was 15 years old. Throughout the conduct and writing up of the review, she advised on the aims and objectives, reminding us of the family perspective and emphasising the need for the review to be relevant to the needs of children and families. She acted as a co-author, revising the final draft of the systematic review for clarity before submission for publication. Her mother, Najma, supported her to do this.

3.3. Specialist Paediatric Palliative Care: What are the benefits? A systematic review

Introduction to the review

Specialist paediatric palliative care services are defined in UK and European standards as those supported by a physician with specialty training (a consultant) in paediatric palliative medicine (13, 19, 20). However, paediatric palliative medicine is a relatively new medical sub-specialty, and few countries have doctors trained to this level (23). As a result, there is a tension between this standard of care and the many existing services that specialise in the provision of paediatric palliative care but

lack the support of specialty trained physicians. Current inequities in specialist paediatric palliative care provision and resource, coupled with the increasing pressure on other healthcare services which have traditionally played a key role in palliative care, such as community nursing services and primary care (88-90), are causing inconsistent delivery of palliative care to children. As the number of children who are living with life-limiting and life-threatening conditions rises (7), there is a pressing need to consider how to improve the delivery of palliative to all children who could benefit from it.

Objective

The objective of this systematic review was to examine specifically specialist paediatric palliative care, defined as a palliative care service supported by a physician with specialty training in paediatric palliative medicine, and address the question "what are the distinct benefits of these specialist services to children with life-limiting and life-threatening conditions and their families?" The review also provided an opportunity to identify evidence gaps for further research.

Design

The Centre for Reviews and Dissemination (CRD) guidance for systematic reviews in healthcare, and the Cochrane Collaboration's handbook for systematic reviews of interventions informed the review's methodology (91, 92). The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) Guidelines informed the structure of the review (93). A protocol was registered and published on the PROSPERO database (https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=50677).

Search strategy

Information sources: A search of the following electronic databases was conducted between September 2015 and January 2016 with the last search on 07.1.2016. The search was repeated in April 2019.

- Cochrane Central Register of Controlled Trials and Cochrane Database of Systematic Reviews
- PubMed (1980 onwards)
- EMBASE (1980 onwards)
- CINAHL (1981 onwards)
- AMED (1985 onwards)

After initial broad scoping searches, the search terms outlined in table 3.1 were developed. The systematic search started with the population search terms, followed by the intervention search. Hand searching of references, "cited by" and PubMed related articles link searches followed. The University of Warwick specialist librarian provided advice on the search strategy.

Table 3.1: Systematic Review Search Strategy

Population	Pediatr* / Paediatr*
	AND (Infant OR Child* OR Adolescen*)
Intervention	Palliat*
	Special*
	End-of-life
	Hospice
	Terminal care
	Consultant
	Physician
	Delivery of healthcare
	Service

Inclusion / Exclusion Criteria

Table 3.2 provides detail of the inclusion and exclusion criteria (92, 94).

Table 3.2: Systematic Review Inclusion and Exclusion Criteria

PICOS Dimension	Inclusion	Exclusion
Population	Children and young people aged from 0-18 years (inclusive)	Studies concerning neonatal palliative care
		Studies concerning specifically young people making the transition to adult services
		Adult studies
Intervention	"Specialist Paediatric Palliative Care" defined as a palliative care service supported by a physician with speciality training in paediatric palliative medicine, as per current recommendations and service specifications.	Paediatric palliative care services that did not meet the specialist specification, including hospice services, and services supported by paediatricians who had not received specialty training in paediatric palliative medicine (where it was possible to establish this).
		Neonatal palliative care services
		Adult palliative care services (who may be catering for paediatric patients)
		Any other usual care

Comparator	Usual care or palliative care that was provided by other types of service	
Outcomes	Any formal measure of evaluation concerning the acceptability or effectiveness of the intervention.	
Study design	Any evaluative study design	Review articles, descriptive or theoretical papers that did not present original research findings
Publication	Databases were searched from 1980 onwards. Unpublished grey literature	Non-English language papers Articles only available in abstract form where no full text is available (the authors were contacted) Voluntary sector reports

Study selection: Duplicate articles were removed. Title and abstracts were screened, followed by examination of the full text. Three reviewers (SM, KB and AM) independently assessed the articles for inclusion.

Data management: Two reviewers extracted relevant data to an Excel spreadsheet (AM and KB), which was independently checked for accuracy and detail by SM. The team discussed any disagreements.

Data synthesis: The included studies were compared and contrasted using a data extraction table. There were no comparable statistics and therefore a systematic narrative synthesis (95) was undertaken, identifying crosscutting themes from each study.

Results

Study selection: The initial search identified 770 relevant articles. 755 were excluded after title and abstract screening and the removal of duplicates, leaving 15 articles. Three of these were excluded because they were conference abstracts of ongoing studies that were not available as full text articles or as unpublished studies from the authors. After applying the inclusion and exclusion criteria to the remaining twelve articles, four were excluded because they did not concern specialist paediatric palliative care services with a specialist physician, leaving eight articles.

The search was repeated in April 2019, with a further 149 articles identified, all of which had been published since the initial search. 145 were excluded following title and abstract screening, and

application of the inclusion criteria. These processes are summarised in Figures 3.1 and 3.2. Study characteristics are summarised in a table presented in Appendix 2.

Figure 3.1: Systematic review PRISMA flow diagram for initial search:
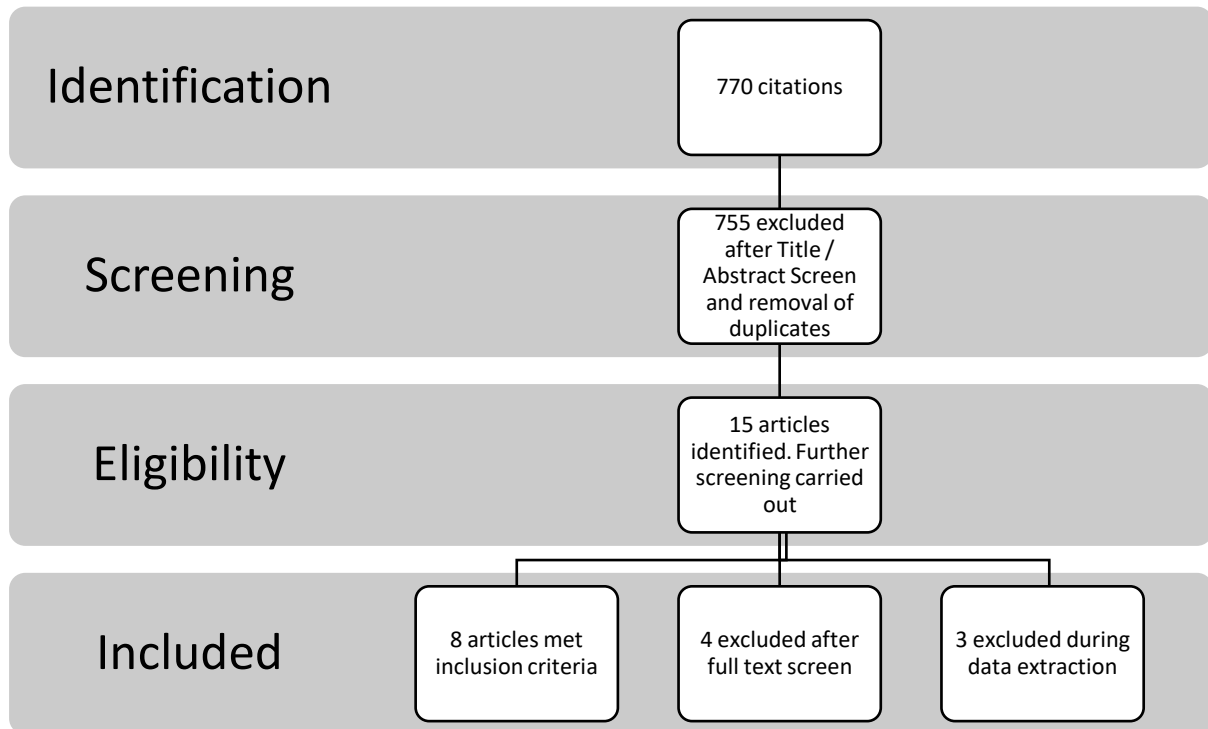
Figure 3.2: Systematic review PRISMA flow diagram April 2019
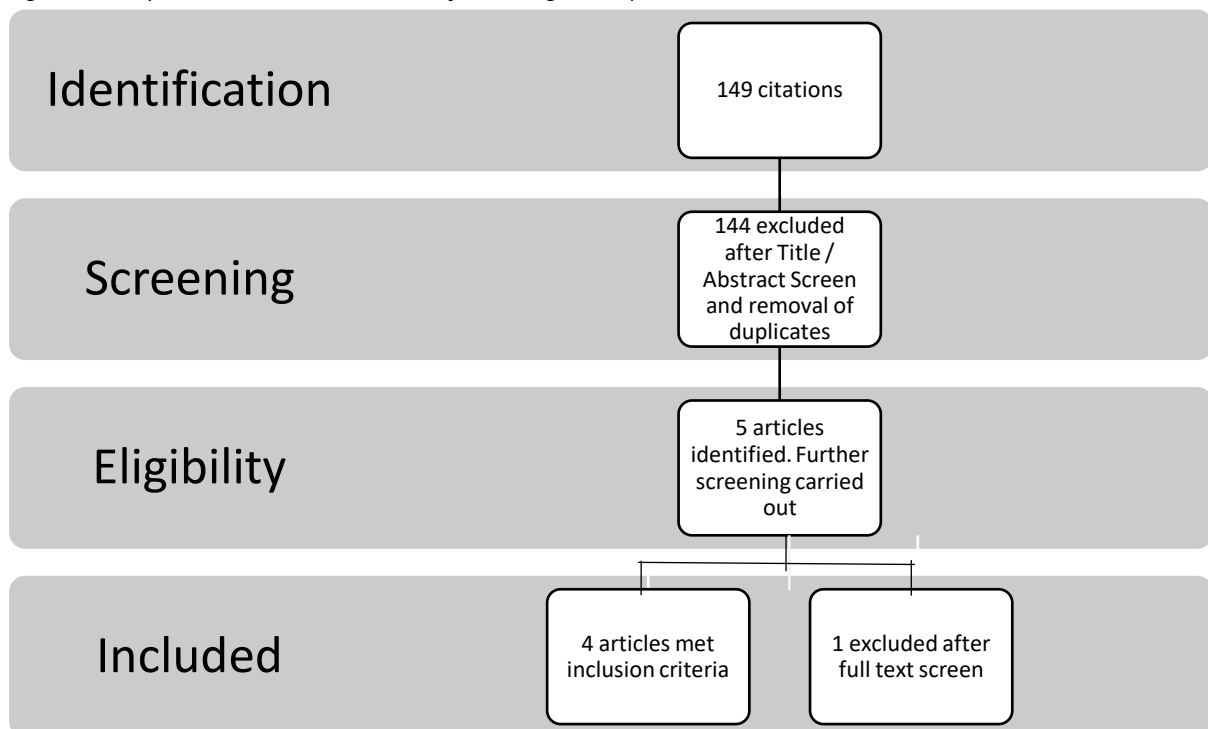
Study location: Included studies were from the USA (96-99), the UK (100), Germany (101-103), Australia (104), Canada (105, 106) and Singapore (107).

Study quality: The studies represented a heterogeneous body of evidence. Three were surveys of bereaved parents (96, 98, 105), and one was a repeated cross-sectional cohort interview study with parents (103). Two were epidemiological studies (100, 106) and four were medical notes reviews (97, 99, 102, 104), one of which included an economic analysis (99). There was one prospective longitudinal survey (101), and one structured impact and cost evaluation of a service (107). There were no randomised-controlled trials or systematic reviews. All had clear aims and used appropriate methodology, and approached the ethical issues. All acknowledged the limitations in their study design and recruitment strategies, and data was collected in a way that would address the research aims. All gave clear descriptions of their data analysis, results and findings, all of which are listed as important in the Critical Skills Appraisal Programme checklist (108).

All of the studies had been published since 2012. The largest study in terms of patient numbers was an epidemiological study, which looked at data regarding 2508 children but was limited by missing data items (100). The notes review studies examined the care of 686 children in total (97, 99, 102, 104). Five studies concerned only children with cancer (96, 100, 103, 105, 106). The other seven studies concerned services for children with non-malignant conditions as well as those with cancer (97-99, 101, 102, 104, 107).

The three surveys of bereaved parents included 200 participants (96, 98, 105). Time since bereavement ranged from 7 months to over four years (96, 98, 105). Response rates for postal surveys of a total number of bereaved parents were 65/192 (37%) (98) and 60/166 (36%) (96). A response rate of 75/140 (54%) was achieved where eligibility criteria were applied (105). The highest response rate for a questionnaire survey was 93% (40/43), with the questionnaire administered face to face with family members at the time they were receiving care from the specialist paediatric palliative care service. This study also attempted the assessment of children by self-report but due to young age and clinical condition this was possible with only three children (101).

Key themes

The four key themes identified about the beneficial impact of specialist paediatric palliative care services on children and their families were:

1. Quality of life
2. Symptom control
3. Place of care
4. Family support

Quality of Life

The studies provided evidence that specialist paediatric palliative care services contribute to improving the quality of life of the child and family through emotional support, care planning and help with medical decision making (101, 104, 107), as well as through the management of distressing physical symptoms (104). Parents reported improved satisfaction with care once specialist paediatric palliative care services were involved (101). One study reported that children who were in contact with a specialist service 'had more fun and [were] more likely to have an experience which added meaning to their life' than those who were not (96).

Symptom control

Pain and symptom management was one of the main reasons for referral to specialist paediatric palliative care services (104). Improvements in children's symptom control with the involvement of the specialist team were reported by parents retrospectively in two studies (96, 101), although perceptions of symptom occurrence, symptom burden and effectiveness of symptom control remained stable over time in another (103). The involvement of specialist paediatric palliative care services resulted in more care, including symptom management, delivered in the home environment, alongside other support for caregivers and practical support (101).

Place of Care

The studies provided evidence to suggest that referral to specialist paediatric palliative care services was associated with fewer admissions to hospital (97, 100, 101, 107), a reduced length of stay (99), and fewer high intensity treatments at the end of life (106). The involvement of specialist paediatric palliative care services was also associated with care planning discussions and opportunity to consider a preferred location of death (105). More children died at home with the involvement of specialist services than not (96, 101, 107). Differences in terms of both diagnosis and geographical location of the family home contributed to the location of death. In one study, children with a cancer diagnosis were more likely to die at home if they lived in a rural location; children with non-malignant disease were more likely to die in a tertiary hospital (104). There was evidence that "goals of care" discussions tended to occur relatively late, with the median time before death that this discussion took place being 16 days (97) .

Family experience

Specialist paediatric palliative care intervention contributed to an improvement in family members' quality of life in a number of different ways (101, 105). Access to services 24 hours a day, 7 days per week was valued (101). Perception of psychological support and support for carrying out "day-to-day activities" increased, and there was a decrease in anxiety and depression amongst parents (101). Specialist teams provided support with medical decision-making (97, 98), including discussions about resuscitation (105), help with communication between family members, including with their child,

and with other healthcare teams (101, 105). The specialist team also played an important role in educating parents about both the process of death and aspects of the medical system (97).

Low referral rates to specialist paediatric palliative care services were described (100) and the average length of time that a child was under the care of the specialist team varied from 20 days to over one year (99, 104, 105). Feedback from families included a desire that specialist paediatric palliative care was involved earlier on in the course of a child's illness (98). In Germany, where legislation requires the provision of specialist services to children with life-limiting and life-threatening conditions, involvement had increased (103).

Discussion

This review set out to investigate the current evidence regarding the distinct benefits to children and their families of care provided by specialist paediatric palliative care services, defined as services supported by a physician with specialist training. A strength of the study is that it focussed specifically on studies that described this model of service delivery. The review questions were informed by PPI with children and young people. The systematic narrative synthesis was conducted by myself, AM and KB, with regular meetings and input from the supervisory team. The study was written up with regular review by a PPI co-author (LS), with feedback provided on relevance to family experience. A small number of studies met the inclusion criteria, all of which had been published within the last seven years. Most were single centre studies with relatively small patient numbers, and rank low in the hierarchy of evidence due to their methodological limitations (109). This is a well-recognised problem in palliative care research, and brings into question the value of systematic reviews in this subject area (110). Specific methodological challenges exist around gatekeeping of participants by clinicians (111), described as a barrier to recruitment in one study (105). In surveys, families were unreachable by phone or post, did not respond, or were ineligible to participate due to language barriers (96, 98, 101, 105). Where families did respond, they were likely to have been particularly motivated to participate, and therefore the survey findings may not have been generalisable to a more diverse population of families. One study tried to collect the views of children, but found this was not possible (101).

Benefits of specialist paediatric palliative care services
Despite the limited quality of the evidence, there were crosscutting themes from the 12 studies suggesting that specialist paediatric palliative care services enable improved quality of life for children and parents, improved symptom control, have an impact on the place of care and an increased likelihood of achieving a preferred place of death. There is also emerging evidence to suggest that the provision of specialist paediatric palliative care is associated with improved

resource utilisation (107), and the delivery of fewer high intensity treatments to children at the end of their lives (106).

Service delivery
The review is timely as the specialty of paediatric palliative medicine further develops, with a range of international standards, specifications and recommendations (20, 21, 112, 113). Systematic consideration of the available evidence to support the development of, and investment in, new services is necessary, particularly as the number of children with life-limiting and life-threatening conditions continues to rise.

There is ongoing emphasis on place of death as an outcome measure in palliative care, despite a limited evidence base to support this (114). Research evidence suggests that families want the option of care provided at home (33), but defining a preferred place of death is a complex issue. Some families value the extra days of a child's life provided in clinical environments such as the paediatric intensive care unit (53). This review suggests an association between care provided by a specialist paediatric palliative care team and opportunity to firstly express preferences for "goals of care" and location of death, and then achieve these (96, 97, 105), although there was some evidence that this occurred relatively late in the course of illness (97). Key factors that enable these discussions are continuous relationships, time for open, honest conversations, and the provision of symptom control (33, 96, 101, 115).

Parents did not always perceive adequate control of their child's symptoms (96, 98), but there was evidence to suggest that more strategies to achieve effective symptom control could be delivered in the home environment, rather than hospital, when the specialist paediatric palliative care team were involved (96). Further research into symptom management in children including use of medications and routes of administration, both in community and inpatient care settings, is an important focus for specialist palliative care innovation and future research.

Specialist paediatric palliative care service design
What cannot be ascertained from the available evidence is which elements of specialist paediatric palliative care services are directly associated with the benefits described, the mechanisms by which these benefits occur or the impact of the presence of a specialty-trained physician. This review looked specifically at services with a specialty-trained physician, and excluded studies of other models of care. However, nurse-led paediatric palliative care services and children's hospice services also provide benefit to children and their families particularly in terms of place of care (116-119), co-ordination of care (109) and family support (120). Research to compare the different types of services would be valuable. Further research to investigate the most effective services for children

with differing diagnoses would also be of value, given the wide variation in disease trajectories, healthcare requirements and family need (121). The implementation of new policies and guidance, including the NICE guidance published in 2016 (20) should be accompanied by robust plans for evaluation.

The benefits of a specialist physician in a service have been broadly described as advanced clinical expertise, and academic, educational and strategic leadership (122), all of which are important in specialist paediatric palliative care as the specialty develops further. Securing funding to develop both specialty training and new consultant posts presents a major challenge and requires the development of clear business cases. The research included in this review provides evidence to support future investment. Raising the profile of specialist paediatric palliative care within healthcare organisations, which traditionally place focus on cure-orientated medical management for children, requires strategic leadership and increased understanding of the benefits of this specialist care. In time, larger, more established teams, with the ability to provide education and training, have the potential to raise the profile and benefits for children and families even further. Research and evaluation should support future innovation and development of the specialist paediatric palliative care workforce.

This review identified only one study that referred to the value of parental input into the development of future specialist paediatric palliative care services (98). Co-design of services with children and families (43, 58, 123), and work to address possible reasons for low referral rates to specialist services, such as negative perceptions of palliative care amongst families (124) and healthcare professionals (18) would be highly relevant.

Conclusion

Future recommendations for service development should address the need for accessible and sustainable specialist paediatric palliative care services for all children who need them. However, as this review demonstrates, there is currently limited evidence to inform policy guidance within the overall provision of paediatric palliative care. In the context of a growing number of children and families who could benefit from palliative care, there is a need for further research, innovation and debate. Robust evaluation of services, care models and professional roles, as well as research to understand how benefits are achieved for children and families, are necessary. These are key considerations for those who are leading the development of specialist paediatric palliative care, and for service commissioners.

3.4. Achieving beneficial outcomes for children with life-limiting and life-threatening conditions receiving palliative care, and their families; a realist review

Background to the review

The systematic review has brought together research evidence that suggests that the involvement of specialist paediatric palliative care services in the care of children and young people with life-limiting and life-threatening conditions is associated with a range of benefits. These include improved symptom control and quality of life for children, their family members feeling more supported, a greater likelihood of care in a place of the family's choice (4), fewer emergency hospital admissions (107), and fewer intensive care treatments at the end of life (106). However, there are significant inequities in the funding and provision of these services internationally, so specialist services do not have the capacity to manage every child who could benefit from palliative care (13, 23, 38), and there are marked inconsistencies in how children and their families experience such care. Outcomes described as important in policy, including Advance Care Planning (ACP) and discussions about a preferred place of death, are not consistently offered to children with life-limiting and life-threatening conditions and their families, and the evidence base to support those interventions is limited (114, 125).

Rationale for a realist review

Realist review is a theory-driven, explanatory, systematic approach which aims to investigate how, when, who for, and to what extent a particular intervention (or "programme") works (126, 127). Realist review of the evidence relating to paediatric palliative care has the advantage over other review methods in that it allows for detailed consideration of palliative care as a broad and complex intervention. It takes into account the fact that palliative care requires the active input of individuals, specialists and non-specialists, who are embedded in social infrastructures such as hospitals and community services, and whose roles are influenced by others, including patients and colleagues. The impact of institutional and system factors, such as local and national policy guidance and commissioning, provide further complexity.

The goal of a realist review is to explain the contexts (C) in which hidden underlying mechanisms (M) are triggered in order to generate outcomes (O) of interest. Context-mechanism-outcome configurations (CMOCs) are proposed and used to develop a programme theory that is 'useful', 'testable' and policy relevant (86). Table 3.3 provides a glossary of realist terms:

Table 3.3: Glossary of realist terms (Adapted from Papoutsi et al (128))

Term	Explanation
Context	Pre-existing structures, settings, environments, circumstances or conditions that influence whether or not certain behavioural and emotional responses (i.e. mechanisms) are triggered.
Context-Mechanism-Outcome Configurations (CMOCs)	Describe the causal relationships between contexts, mechanisms and outcomes i.e. how certain outcomes are achieved through mechanisms being triggered in certain contexts.
Mechanisms	The behavioural or emotional response which is triggered in certain contexts. Mechanisms are context sensitive and are usually hidden.
Outcome	The impact of mechanisms being triggered in certain contexts.
Programme theory	A set of theoretical explanations about how a particular programme, process or interventions is expected to work.
Mid-range theory	Theoretical explanations which are suitable for testing through further research. A programme theory can be specified at the mid-range.

Initial programme theory

As outlined in the introductory chapter of the thesis, the initial programme theory, drawn from research and policy, is that palliative care for children "works". The systematic review has provided evidence to refine that theory, describing the benefits experienced by children and families when specialist paediatric palliative care services are involved in their care.

Aim of the review

This realist review aims to examine palliative care more broadly, and describe when, how and in what circumstances palliative care provides benefits from a child and family perspective. The realist approach allows the description of CMOCs and the proposal of a programme theory that will add to the understanding of how palliative care can be delivered more broadly as an approach to care for children and families, and so form the basis of policy relevant recommendations.

Methods

The review was conceptualised in August 2015 and carried out over the following two years. Ethical approval was not required. A protocol was published in PROSPERO (registration number: CRD42018090646 https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=90646).

The review followed Pawson's five stages for a realist review, and the RAMESES standards (129). The first stage was the identification of the initial programme theory, and clarification of the purpose of this review (i). A detailed iterative search for research evidence followed (ii). Articles were selected for inclusion based on their relevance to the research questions (iii), and relevant data were extracted and organised into a Word table (iv). The final stage of the review was data synthesis; developing CMOCs and a testable, mid-range programme theory (v) (127, 129).

Step (i): Identification of initial programme theory and clarification of the scope of the review

The initial programme theory, that palliative care for children "works", was informed by policy documents (as outlined in Chapter 1), and the systematic review. A scoping review was conducted, comprising an exploratory internet based literature search, review of policy documents, the collection of relevant articles via social media and at conferences (Table 3.4), and regular discussion with a stakeholder group of professionals and parents (the West Midlands Paediatric Palliative Care Network) who met every three months through the course of the review.

Table 3.4: Sources of information to identify existing theories

Area of initial search	Sources
Internet	Google, Google Scholar, NHS, voluntary sector and government websites, and the Cochrane library
Desk-drawer search	Articles already known to the researchers Search of key textbooks
Social media	Saving relevant articles found through Twitter, Facebook and Together for Short Lives Newsletters
Conferences	Posters and presentations, abstracts Reflective notes
Stakeholders	West Midlands Paediatric Palliative Care Network meetings Reflective notes

The scoping review revealed a diverse range of literature in paediatric palliative care, with articles focussing on many different aspects of care including the child and family experience, symptom control, advance care planning, organ donation, complementary therapies, spirituality and the perceptions of healthcare professionals. Following discussion with the stakeholder group and research team, the research team made a decision to focus on the experiences of children and their families in relation to palliative care, prioritising research that provided insights into their experiences and perceptions, rather than the experiences of professionals. The research questions that emerged were as follows:

1. What are the beneficial outcomes (O) described by children with life-limiting and life-threatening conditions and their families in relation to palliative care?
2. What are the mechanisms (M) by which these beneficial outcomes are achieved?
3. What are the contexts (C) that determine whether these mechanisms produce the outcomes?
4. What are the implications for future research, policy and practice?

Step (ii): Systematic literature search

A formal database search was designed with support from the specialist librarian at the University of Warwick, and the search carried out in November 2015. Broad search terms were tested in PubMed (Palliat* AND Paediatr*/ Pediatr*); searches were then carried out in AMED, ASSIA, CINAHL, EMBASE, PsychINFO, Web of Science and ERIC, with the search terms modified and adapted for each database, but kept deliberately broad. Forward and backward citation tracking was conducted. The database search was of papers published since 1980, but no articles were excluded based on date of publication. The search was limited to papers published in English. Relevant references were collected over two years via citation alerts, social media and at conferences, and the database search repeated in December 2017. The aim was to gather evidence to refine and test the initial programme theory, rather than to conduct an exhaustive search of the paediatric palliative care literature.

Step (iii): Document screening and selection

References were exported to citation management software (EndNote), where screening for duplicates was carried out. All of the titles and abstracts were reviewed in chronological order, to gain an understanding of the shifts and changes in the literature over time. The articles were grouped into categories according to the subject and focus of the research. Table 3.5 outlines the inclusion and exclusion criteria. Articles that provided empirical research evidence or family accounts about the experiences of children and families in relation to palliative care that would inform the programme theory were included and retrieved as full texts. Expert professional opinion articles, practice reviews and editorials were deliberately excluded.

Table 3.5: Inclusion and exclusion criteria for the realist review

Inclusion	- Empirical research or systematic reviews about the experiences of children and families in relation to the delivery of palliative care, either Specialist Paediatric Palliative Care Services (those supported by a consultant in Palliative Medicine), other paediatric palliative care services, or any important aspect of palliative care such as communication. - Children and/or families are the research participants - Children are defined as 0-25 years of age (palliative care services and research studies vary in their age thresholds) - Children with life-limiting or life-threatening conditions (as defined by Together for Short Lives (2)
Exclusion	- Opinion pieces, editorials and practice reviews - Research about the opinions and experiences of healthcare professionals - Neonatal / antenatal / adult palliative care

Step (iv): Extracting and organising data

The review team (SM, KB and AM) read and re-read the articles, and met regularly to consider the trustworthiness and rigour of those that were included. Article characteristics (citation, year, and country, type of paper, aims, methods and participants) were summarised in a Word data extraction table (Appendix 3) by KB and AM, with regular discussion and consistency checking with SM and the research team. SM and KB coded relevant sections of text, using a process of manual annotation and data management software (NVivo). An inductive approach was taken, with codes and concepts originating from the data, using the following questions to guide the process (130):

1. What does this section of text describe about the important factors in relation to palliative care for the child and family?
2. Is the section of text referring to context, mechanism or outcome?

A second data extraction table (provided in Appendix 4) was used to document key relevant sections of text that were used to inform interpretations about what was functioning as context, mechanism or outcome within CMOCs.

Step (v): Data analysis and synthesis

The aim of the data analysis was to interpret and explain the "hidden" mechanisms, triggered in certain contexts, such that palliative care can lead to beneficial outcomes for children and families. Coded sections of text informed the development of CMOCs, using the following questions as a guide:

1. What is the context? What outcomes are described? What are the hidden mechanisms? What is the CMOC?
2. How does the CMOC relate to patient and family experience?
3. Is the evidence trustworthy and rigorous?

SM and KB conducted the analysis, with consistency and accuracy checking, and discussion of potential CMOCs with the wider research team (SM, JD and A-MS). Analytical strategies included juxtaposition of data sources (aligning evidence to inform and clarify a theory), exploration and reconciliation of discrepancies in the data and adjudication of data quality (129, 131). Where further evidence was required to adjudicate an argument, SM conducted a purposive search for further data, in the organised dataset from the wider literature search (stored in EndNote).

Search Results
The database search took place over several months. 5,930 articles were identified (Table 3.6).

Table 3.6: Realist review literature search results

Database	Search terms	Articles found on searching	Articles selected after title and abstract screening
AMED	Palliat* AND (Paediatr* or Child*)	721	209
ASSIA	Palliat* AND (Paediatr* or Child*)	643	29
CINAHL	Palliative care AND paediatric / children	168	41
EMBASE	Palliat* AND Paediatr*	1041	140
PsychINFO	Palliat* AND Paediatr*	69	28
PubMed	Palliat* AND Paediatr*/ Pediatr*	1805	181
Web of Science	Palliat* AND Paediatr*	1339	89
ERIC	Palliative care AND Paediatric / children	144	2
Desk drawer search	N/A	55	55
TOTAL		5930	719

55 further articles were identified through desk drawer searching, forward and backward citation searching and the collection of articles from social media. 5,211 were excluded after title and abstract screening, as they were either not relevant to the research questions or were duplicates. 774 articles were grouped into broad conceptual categories according to the focus of the research. 714 articles were editorials, opinion pieces, practice reviews and research that did not include children and families as participants. 60 articles met the inclusion criteria (children and families as the research participants) and comprised the final data set. The children included in the studies had a diverse range of life-limiting and life-threatening conditions. The table in Appendix 3 provides the characteristics of the included studies. The PRISMA flow diagram shows the data screening and extraction processes (Figure 3.3) (93).

Figure 3.3 Realist review PRISMA flow diagram

Database search	5930 citations (719 after title and abstract screening)
Forward and backward citation searching, desk drawer search and social media links	55 further citations
Citations selected for further screening	774 citations
Focus on child and family experience	60 citations
Included	51 original research papers / 4 family narratives or case studies / 5 literature reviews

Of the research papers, 51 were original research, five were literature reviews (132-136), two were first person family narratives (137, 138), there was one case study (139) and one analysis of a diary (140). 22 studies included children with an oncology diagnosis (115, 132, 133, 136, 137, 139-155) five concerned those with non-malignant disease (138, 156-159), and 33 included both (27-29, 33, 51, 98, 106, 134, 135, 160-183).

Two research studies included siblings (174, 177), 15 were carried out with parents (28, 29, 144, 148, 149, 152, 156, 164, 165, 168, 169, 172, 175, 180, 181), one included parents and grandparents (157), and four were carried out with both children and parents (33, 162, 167, 170). Three studies included children only as participants; a retrospective cohort population study (106), one qualitative interview study where children were interviewed alone (154), and one longitudinal observational study (158). The other 26 research studies were carried out with parents post-bereavement (51, 115, 141-143, 145-147, 151, 153, 155, 159-161) (27, 98, 163, 166, 171, 173, 176, 178, 179, 182, 183); one also included siblings (177).

Studies were heterogeneous in terms of methods; the majority made use of qualitative methods including individual interviews (29, 33, 51, 115, 141, 143-146, 148, 151-156, 160-164) (27, 166, 170, 173-175, 180) (28, 182), focus group interviews (33, 147, 150, 171, 180), or written questionnaires (98, 142, 159, 170, 176-178, 181). Several studies conducted quantitative analysis on questionnaire findings (115, 165, 168, 172, 173, 179). The studies represented an international evidence base, with

studies from the UK (27, 33, 137, 138, 147, 155, 167, 170, 172, 180), Australia (28, 29, 145, 146, 176), Canada (106, 148, 149, 151, 156, 157), and the USA (51, 98, 115, 133, 134, 136, 139, 141, 158-166, 168, 169, 171, 173, 175, 181), Germany (143, 150), Holland (152, 178, 179) India (144), Ireland (132), Malaysia (182), New Zealand (174, 177), Sweden (135, 142, 153, 154), Switzerland (183), and Turkey (140).

Findings:

The review findings provide insights and understanding into the beneficial outcomes described by children with life-limiting and life-threatening conditions and their families in relation to palliative care, and when and how these are achieved. Four conceptual areas were identified: (i) family adaptation and experiences, (ii) the child's situation, (iii) relationships with healthcare professionals, and (iv) access to palliative care services. A narrative is provided for each area, followed by realist analysis and CMOCs. A programme theory, derived from the CMOCs, is then presented.

1. **Family adaptation and experiences**

A child becoming seriously unwell or dying alters family life in ways which parents and siblings cannot anticipate or prepare for (144, 145, 157, 164, 174). Parents grieve for the loss of the child's health, struggle with a feeling of responsibility for their child's wellbeing and have to adjust their hopes and expectations of parenthood and the future (142, 144, 164, 168, 180). The diagnosis of a condition such as cancer brings an immediate realisation of the precariousness of life (145, 157), whereas parents of children with non-malignant, congenital conditions describe a more gradual realisation, with the severity of the child's condition being under-emphasised by healthcare professionals who are "too considerate"(29).

Families adapt over time, carrying out essential practical tasks (33, 172) and becoming experts in both their child's condition and the impact it has on their family (33, 132, 137, 169). They find new meaning and purpose in their lives (153, 157, 169), adopting the role of carer, spending more time in hospital and leaving work, which can lead to feelings of vulnerability, isolation, fatigue, depression and anxiety, and a perception that no-one understands the family's burdens (28, 168, 169). They draw upon support from a wide variety of sources including other parents of children with the same condition, friends and the local community (153, 156, 164, 168, 178).

Life with intensive medical treatments and chronic uncertainty becomes normal (27, 139, 157, 159) and the parent-child bond develops in the context of an illness that is often characterised by unexpected crises and "moments of realisation", when the threat to the child's life is recognised (28, 160, 167, 168). Coping with this normality is challenging and stressful (172, 181). Parents adopt a number of strategies such as trying to maintain hope and "staying positive" (147, 148, 154, 157,

160). Parents and families describe a need to be respected as experts in their child's condition, to be involved in care decisions and for their beliefs and opinions to be taken seriously at times when their child is critically unwell and may die (134, 149, 163, 165, 166, 175, 177, 179), but this does not always happen in practice (173). As "protectors" of the child (28), parents are caught between conflicting emotions, neither wanting their child to suffer, nor wanting their child to die (153), but they may not have to fully acknowledge that their child is dying in order to be willing to place the emphasis of care on lessening of suffering (141). When difficult decisions are to be made, affirmation in their decision-making from a healthcare professional who has witnessed the magnitude of the task is valued (176).

Parents can experience disempowerment related to the healthcare environment in which their child is receiving care. The intensive care unit has been described as "bewildering" (51), and parents have described feeling unable to raise concerns about their child's care if they feel grateful to a service or perceive that by virtue of being in a specialist centre, their care is the best it can be (115, 160, 170). Clinical concerns, including symptoms, have been found to be under-reported by healthcare professionals compared to parents who may not always feel able to raise their concerns (115, 142, 156).

Studies suggest that healthcare professionals recognise that a child is dying before family members do (115, 141). This may happen very late in the course of illness, sometimes not until death is imminent (159). Parents describe receiving the news that their child is going to die as "a crushing, stunning defeat after a prolonged and painful struggle" (137), like "gripping my heart and squeezing" (155) and "like being covered in a wet and dark blanket" (153). They may have difficulties understanding and assimilating information about the incurability of their child's condition (153), perhaps because this represents a significant change from a cure-focussed management plan, particularly when the underlying condition is cancer. Some parents are never explicitly told that their child is dying (173). In contrast to studies of healthcare professionals, family narratives and case studies suggest that family members are aware of the possibility of the death of the child throughout the course of illness (138, 139, 155, 160, 167).

Realist analysis:
There is much to learn from the literature about the experiences of families when a child has a life-limiting or life-threatening condition. Descriptions of their experiences highlight important contexts for the delivery of palliative care, as both a broad approach and as a specialist service. These contexts include the fragility of the child's condition and chronic uncertainty. Mechanisms triggered in these family contexts include adaptation to a situation that is against cultural norms, continually

adjusting expectations for family life and developing coping strategies (mechanisms). Family members frame and re-frame their hopes and expectations (mechanism) and develop significant expertise in the management and impact of the child's condition (outcome). Families are disempowered and intensely vulnerable in their situation, both in terms of the uncertainty that they live with, and in their interactions with healthcare environments and systems (context). They have an awareness that their child may die, but this may remain unspoken until late in the child's illness (mechanism). However, this awareness may allow them to place the emphasis of care on lessening suffering (outcome), even if the possibility of dying remains unspoken. These CMOCs are outlined in Figure 3.4 below:

Figure 3.4: CMOCs relating to family adaptation
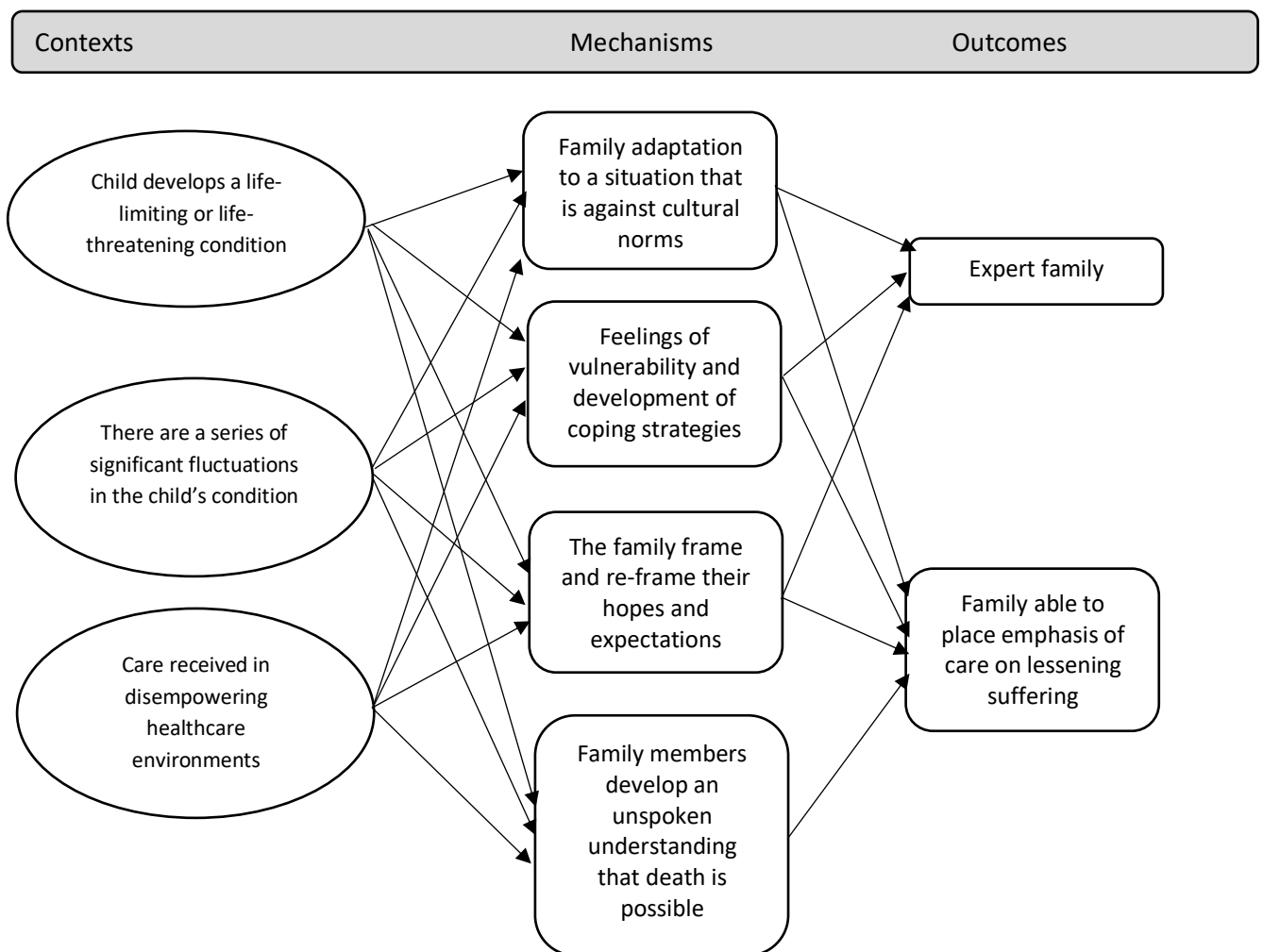
2. **The child's situation**

The ability of children with life-limiting and life-threatening conditions to take part in conversations about their healthcare varies according to their age, developmental stage, psychological and

cognitive factors related to their condition, and the behaviour of the adults around them (173).
Parents are often the surrogate decision makers, with children becoming passive recipients of the
decisions that are made for them (115), a situation in which they display both resilience and dignity
(137).

In the few (seven) studies where children participated, they expressed a desire to live their lives as
normally as possible despite their abnormal circumstances (33, 147). Their priorities included seeing
friends and attending school (170). They wished to receive truthful information, in a way that they
could understand and at the same time as their parents (154).

Parents worry about a right or wrong way to discuss death and dying with their children (137, 174).
Cultural beliefs, a desire to protect the child, or a perception that their child is ambivalent about
taking part in healthcare discussions lead parents to consider conversations with their child about
the possibility of death to be inappropriate or unacceptable (144, 171, 174). Even without
conversations, parents describe seeing their child's understanding of their situation change over
time (139), as they develop a "tacit understanding" that they may die. Some parents and caregivers
feel that explicit conversations about dying become unnecessary because the child already
understands the reality of their situation (145, 174).

Realist analysis:

Figure 3.5 outlines the CMOCs related to the child's situation. Children express their own interests
and priorities for life (context); parents are often their surrogate healthcare decision makers
(context). Children may be ambivalent about decisions related to their health, or may be protected
by their parents, therefore becoming passive recipients of the care decisions that are made for them
(outcome). The possibility of dying may not be openly discussed (outcome), but a tacit
understanding that the condition may lead to death has been described amongst children
(mechanism).

Figure 3.5: CMOCs related to the child's situation

Contexts	Mechanisms	Outcomes

- Child has own interests and priorities
- Parents become surrogate decision makers

- Child perceived to be ambivalent towards healthcare discussions
- Parents and child develop a tacit understanding that death is possible
- Parents have a desire to protect the child and fear "getting it wrong"

- Child is a passive recipient of care
- Death is not openly discussed
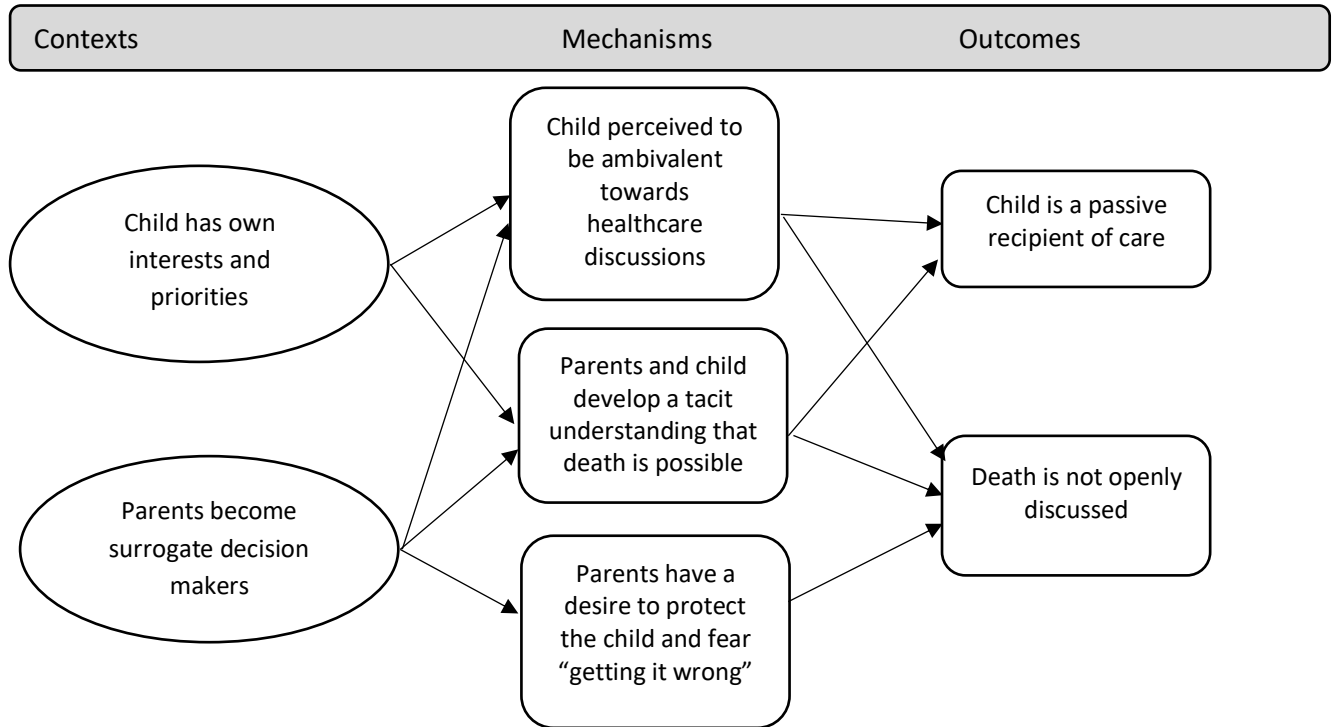
3. **Relationships with healthcare professionals**

The relationships that develop between children, family members and healthcare professionals are critical to the family experience (27, 133, 160). Families describe the necessity of authentic relationships, and want to feel that healthcare professionals are experienced, competent and can be trusted (29, 51). Open, honest communication, care co-ordination, accessibility and availability are valued (146, 151, 160, 162, 171, 173, 181, 183). Children and parents appreciate healthcare professionals who take the time to get to know the child, even to the extent of "developing a friendship" (162). The individualised and intimate knowledge of the family situation that underpins these relationships is often achieved through continuity of care (33, 170). It may be one specific healthcare professional who advocates for the family and is perceived to be particularly helpful (33, 179).

Families value the emotional investment made by some healthcare professionals, demonstrated through compassion and acts such as appearing to care for the child as "one of their own", attendance at a memorial service or making contact in bereavement (29, 51, 161, 171). Being with families at their most vulnerable time requires understanding of the physical and psychological distress that they might be experiencing and an ability to bear this with them, a situation which can lead healthcare professionals to experience their own feelings of distress (98, 164, 173).

Open, honest communication, care co-ordination, accessibility and availability are valued (146, 151, 160, 162, 171, 173, 181, 183). Children and parents appreciate healthcare professionals who take the time to get to know the child, even to the extent of "developing a friendship" (162). Continuity of care is key to achieving the individualised and intimate knowledge of the family situation that underpins these relationships (33, 170). It may be one specific healthcare professional who advocates for the family and is perceived to be particularly helpful (33, 179).

Conversely, relationships perceived as "poor" by parents carry significant risks of harm. A single event, such as the insensitive delivery of bad news, parents feeling patronised or dismissed, or that their judgement has been disregarded, can lead to lasting distress (51, 139, 142, 156, 160, 162). Lack of continuity leading to different healthcare professionals asking the same questions several times can be "disturbing" (135). Times when parents feel the opinions of healthcare professionals have been "inflicted" upon them, or when their individual needs have been subsumed to standard procedures rather than being listened to, may lead to significant conflict (51).

Realist analysis:

There are two important interdependent contexts for healthcare professionals that trigger mechanisms leading to beneficial outcomes for children and families. Individual professionals differ in their approach, with some more motivated to deliver a holistic approach to care (context). Continuity of care allows the development of detailed knowledge of the child and family situation over time (context). Mechanisms triggered in these contexts include respect for the family circumstances, advocacy and affirmation in decision-making, personal emotional investment, and a capacity in the healthcare professional to bear witness to the family situation. These mechanisms lead to outcomes including trusted, authentic relationships between children, their families and healthcare professionals in which children and families feel respected, heard and supported. They feel that the healthcare professional shares the emotional impact of the child's condition (outcome). Figure 3.6 demonstrates these CMOCs:

Figure 3.6: CMOCs outlining relationships with healthcare professionals
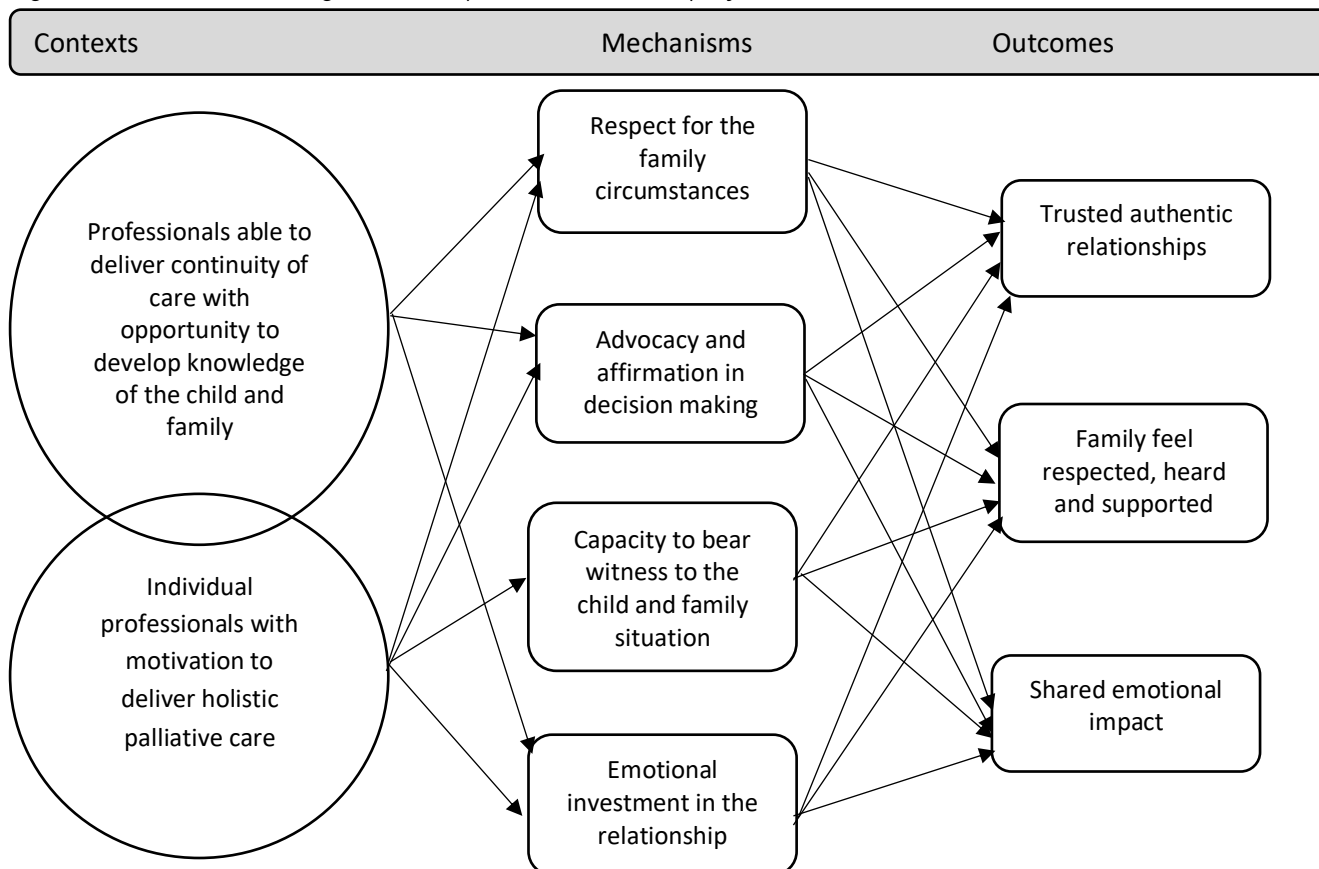
4. **Access to palliative care services**

When available, specialist paediatric palliative care services are associated with a range of benefits including a feeling of support for families and improved symptom control (106, 115, 143, 150, 172, 182). Symptom control can be particularly challenging given each child's individual condition and circumstances (156, 158, 173). However, barriers to referral exist, including variable perceptions and opinions of the term "palliative care" amongst professionals (18), children and their families (124). Research suggests that family members view "palliative care" as a distinct phase at the end of a child's life, "the beginning of the end". They fear it as a point at which they will lose contact with the healthcare services they know, a situation that can be "terrifying" (180).

Parents who receive care from specialist paediatric palliative care services report that they had been introduced to these services earlier in the course of the child's illness (170). They are more likely to accept a referral once they have been provided with detailed information which addresses their own preconceptions of "palliative care" (124).

Children's perceptions of palliative care services are largely unknown. They have been found to be reluctant to accept new services or healthcare providers who are introduced towards the end of life

(171). However, bereaved parents are more likely to describe their children as calm and peaceful during the last month of life if they have had contact with a hospice (115).

Realist analysis:

The analysis so far highlights the intense vulnerability of families who are experts in the care of the child and their condition, when they realise that their child may die (context). The relationships with trusted healthcare professionals that have been established through the course of the child's illness are key and function as a context for the delivery of palliative care, including being able to place an emphasis of care on lessening suffering, and making a referral to specialist paediatric palliative care services (outcome). These are important precursors to being able to consider policy outcomes in the care of individual children and their families, such as advance care planning, and access to specialist palliative care expertise and services. Negative perceptions of palliative care and challenges with introducing new professionals or services late in the course of the child's illness can make the introduction of specialist services difficult as the child approaches the end of their life. The underlying mechanisms, including advocacy, trust, and affirmation in decision-making can all help with this process. These CMOCs are outlined in Figure 3.7 below:

Figure 3.7: CMOCs related to palliative and end of life care
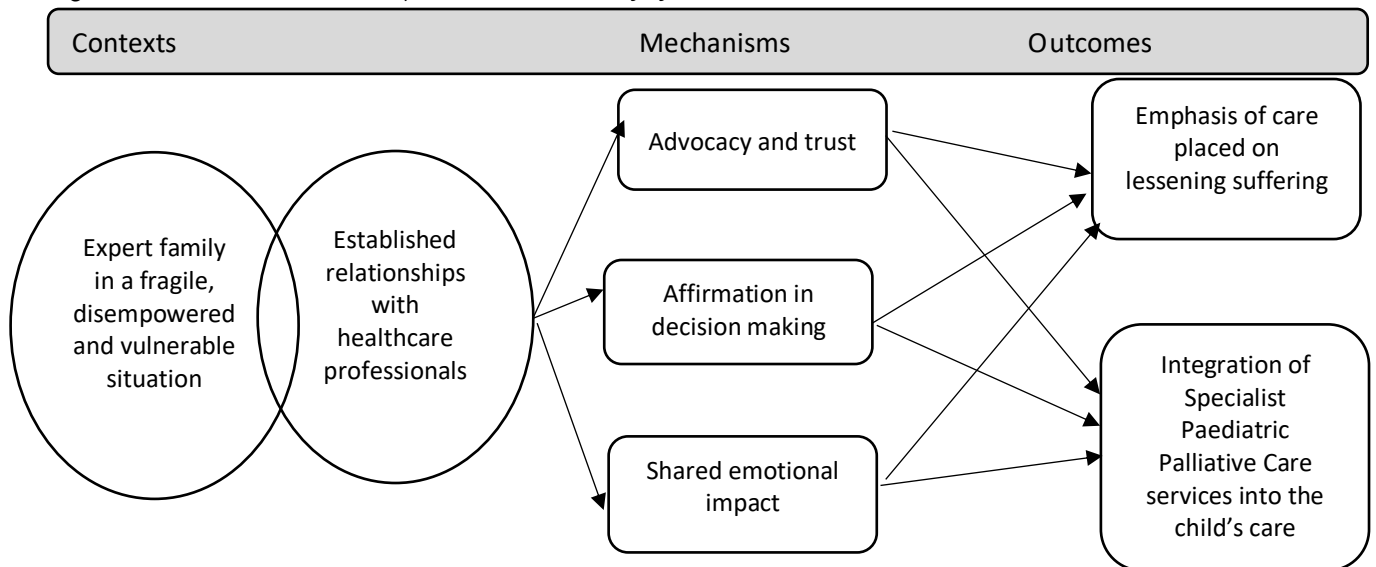
5. **Development of a programme theory**

The realist analysis related to the delivery of palliative care service and policy outcomes starts by taking the outcomes described through the formulation of CMOCs related to the family experience and the child's situation as important contexts for the delivery of palliative care. Firstly, there is a child with his or her own interests and priorities, and secondly, an expert family who are

disempowered and vulnerable in their situation. Both may have an unspoken awareness that the death of the child is possible.

Important child and family-related outcomes are feeling respected, heard and supported, and being able to place emphasis on lessening the child's suffering. These depend on established, trusted relationships with healthcare professionals who are motivated to deliver a palliative care approach and can provide continuity of care through the course of the child's illness. Relationships of this nature are a professional resource context for the delivery of palliative care. The mechanisms that underpin these relationships are key, including respect for the family circumstances, advocacy, affirmation, an ability in the healthcare professional to bear witness to the child and family situation, and emotional investment in the relationship. Through these relationships, outcomes can be achieved including shared emotional impact and open acknowledgement of the fragility of the child's condition and the possibility of dying. These are key precursors to conversations during which child and family preferences and priorities, and referral to specialist paediatric palliative care services, can be discussed (outcomes). Achieving these outcomes would support more consistent delivery of the service outcomes identified in the systematic review. These included improved quality of life and symptom control, and a feeling of support for families. Policy outcomes, including achieving a preferred place of death, are also more likely to be achieved. Figure 3.8 provides a diagram of this proposed programme theory.

Figure 3.8: Proposed programme theory from the realist review:

| Contexts 1: Family situation | Contexts 2: Professional resource | Mechanisms | Child and family-related outcomes | Policy and service outcomes |

Expert family in a fragile, disempowered and vulnerable situation

Child with own interests and priorities

Established, trusted relationships with healthcare professionals

Respect for the family situation

Advocacy and affirmation in decision-making

Ability of professionals to bear witness to the child's and family situation

Emotional investment in the relationship

Child and family feel respected, heard and supported

Emphasis of care placed on lessening suffering

Shared emotional impact

Possibility of death acknowledged

Improved quality of life and symptom control

Integration of specialist paediatric palliative care services into the child's care

Advance Care Planning and preferred place of care discussed

Discussion

Summary of findings

This review has led to the development of a programme theory that proposes how the delivery of palliative care to children and their families could be improved, through a series of explanatory mechanisms, triggered in certain contexts, to produce outcomes described as important to families. The programme theory brings together the contexts, mechanisms and outcomes from the literature and relates these to desired policy and palliative care service outcomes.

The review adds to the evidence base for the development of paediatric palliative care, by providing insights into the highly individual and unique knowledge and experience that families develop about the management of their child with an often complex, life-limiting or life-threatening condition, and considering elements of care required to support them (184). Their hopes and expectations are shaped by constant adaptation to uncertainty and a sometimes unspoken awareness that the child may die. These child and family circumstances are contexts for the delivery of palliative care. Navigation of these complex dynamics requires skill. Previous studies have shown that families seek support from a variety of sources, including other parents (185). Where open, honest conversations with healthcare professionals took place, families had more opportunity to plan their child's end of life care, and achieve outcomes that are consistent with high quality palliative care (186, 187). This review explores the mechanisms triggered when established, trusted relationships with healthcare professionals exist, including advocacy, and emotional investment in the relationship. Child and family outcomes including feeling respected, heard and supported, and a feeling that their emotional burden is shared, can be achieved. These child and family outcomes may lead to a more open acknowledgement of the possibility of death and the ability to place the emphasis of care on lessening suffering.

Strengths and Limitations

The strength of the realist approach is its explanatory nature (188). This review set out to investigate what works for children and families, when, how and in what circumstances in terms of palliative care. The iterative search strategy reduced the risk of missing major concepts that are relevant to the delivery of a palliative care approach to children and families. The evidence included children with a diverse range of clinical conditions, adding to the applicability of the findings across settings. The findings of this review were drawn from the international evidence base identified through the search strategy. The applicability of the findings across different healthcare systems depends on the health service infrastructure. For example, there are differences between the healthcare systems in the UK and the USA, with a more definite referral point to a palliative care service required in the USA due to the nature of funding for healthcare.

67

The majority of the studies reviewed were qualitative, and a strength of the review is that this allowed relevant contexts, mechanisms and outcomes to be abstracted from rich, in-depth data. Given the paucity of research evidence in the field of paediatric palliative care, much current policy to date has been informed by expert opinion. Expert opinion articles, such as editorials and practice reviews, were deliberately excluded from this review, and the focus of the review was on gaining an understanding of the experiences and perspectives of children and families. Personal experience and family narratives published in medical journals were included. The rationale for this was to prioritise evidence related to the child and family experience over expert professional opinion. This approach does however raise a risk of bias, particularly participant bias, with those who are coping or who are more motivated to improve palliative and end of life care for children being most likely to participate in research or provide personal experiences of their accounts in an attempt to influence and effect change. Another possible limitation is that most of the studies included bereaved parents as the participants, with varying lengths of time since their bereavement, and recollections of experiences can change over time (189).

What this study adds
This realist review addresses an important gap in the evidence, providing an understanding of the contexts that are required in order to achieve beneficial outcomes for children with palliative care needs and their families. The insights are valuable given the challenge of translating the words of policy into clinical practice. The programme theory proposes that there are important child and family outcomes, which underpin the delivery of wider policy goals and palliative care service outcomes.

Recommendations for research, practice and policy
In order for policy goals and standards to be achieved in paediatric palliative care, organisational policy and intervention strategies should be developed that recognise the key importance of family relationships with healthcare professionals. Enabling the contexts that trigger mechanisms leading to important child and family outcomes could result in the delivery of a more consistent palliative care approach. Intervention strategies include providing support for those who are motivated to provide palliative care, as well as accessible education and training opportunities. It also requires healthcare leaders and those involved in service design to value continuity of care and to enable time resource for key interpersonal relationships to develop.

Paediatricians are frequently involved in the care of children with life-limiting and life-threatening conditions, and the care of children who die, from early on in their career (190, 191). There is wide variation in the confidence levels of paediatricians in terms of the delivery of palliative care (192), and mixed levels of willingness to undertake further training (192, 193), perhaps because palliative

care as a concept is poorly understood. Accessible and relevant training and education opportunities need to be developed, including increasing awareness and changing attitudes around what palliative care is (18, 194), and education about the role of specialist services, where they are available.

The presence of role models, such as members of a specialist paediatric palliative care team, can have a positive impact in terms of increasing understanding of palliative care (195). Further research to understand how healthcare professionals develop the professional values and behaviours that make the delivery of palliative care possible, including whether there is a "type" of healthcare professional or family that are more likely to engage with palliative care, would be valuable (106).

The provision of clear and comprehensive information to families that outlines available professionals and services, including specialist paediatric palliative care services, early on in the course of the child's condition could potentially be helpful. Currently, they may receive information about available services through informal peer support networks, including via social media. An area for future investigation is the preferred information sources of children and their families, and their needs and preferences regarding that information.

Conclusion

In conclusion, this review has described how outcomes that are important to children and families, including feeling heard and respected, and that their emotional burden is shared, are key to their experience of palliative care. These outcomes are achieved through the development of established, trusted relationships with healthcare professionals, and hidden mechanisms triggered within these relationships including advocacy and affirmation in decision making. Motivation to deliver palliative care, and an ability to bear witness to the child and family situation are necessary within healthcare professionals. These nuanced and hidden influences require more attention, since they lead to child and family outcomes that underpin the standards outlined in policy, such as advance care planning, referral to specialist services and support for families (196).

3.5. Chapter summary

Families face a devastating situation when their child has a life-limiting or life-threatening condition and is at risk of dying. There is evidence from the systematic review that specialist paediatric palliative care services, defined as those supported by a trained specialist physician, are associated with improved experiences for children and families. The evidence base is limited, but it is growing. There is a need to better understand the impact of specialty-trained physicians in paediatric palliative care, since delivery of the model of care outlined in policy guidance (196) is associated with significant training and resource implications for healthcare systems including the NHS in the UK.

The realist review provides in-depth insights into what works, who for and when. The proposed programme theory describes how access to specialist paediatric palliative care services depends not only on resource for these services, but also on the interactions of the child and family with the wider healthcare system. The complexity of the child's condition and vulnerability of the family are highly individual contexts for the delivery of palliative care. Policy makers and those involved in the design of healthcare services including palliative care should recognise the impact and importance of established, trusted relationships between children, families and healthcare professionals. The establishment of these relationships depends on the provision of working environments where it is possible to deliver continuity of care. The delivery of palliative care depends on trust, professionals sharing the emotional impact of the child' condition, and being able to bear witness to the family situation. When these mechanisms are triggered, outcomes that are considered important to families are more likely to be achieved, and could underpin the delivery of services and policy outcomes in palliative care. The role of the specialist paediatric palliative care team is not only in the direct care of the patient, but also in legitimising a palliative approach to care.

The proposed programme theory developed from these two literature reviews requires more testing and refining in order to develop policy relevant recommendations. This is the aim of the empirical research, described in Part 2 of the thesis.

Part Two: Methodology and Methods

4. Methodology

4.1. Overview of Chapter 4

Evidence-based medicine drives policy and clinical practice, and there has been significant emphasis on randomised controlled trials as the gold standard for research. The evidence base in palliative care for children to inform policy and practice is relatively scarce. Randomised controlled trials do not necessarily lend themselves to an intervention as complex as palliative care delivery. Much current practice and policy in children's palliative care is based upon expert opinion, and the need to expand the research evidence base is well recognised.

One of the main aims of this research is to contribute to the evidence base that can inform policy, practice and new approaches to the provision of palliative care to children. This chapter starts with an overview of the potential problems and limitations of applying traditional evidence-based medicine to this area of practice. It goes on to explain the epistemological position I have taken as a researcher, and the established theories that I have drawn upon in order to design the empirical research. The theoretical framework that informs the research is at the end of the chapter.

4.2. Evidence-based medicine and palliative care

In order to explain and justify my epistemological position, and the methodology for the empirical research, it is first important to consider the limitations of more traditional research approaches and evidence-based medicine in palliative care for children.

Evidence-based medicine (EBM), defined as "the conscientious, explicit and judicious use of current best evidence in making decisions about the care of individual patients" (197, 198), was launched over 25 years ago by the EBM working group (199). Heralded as a "new paradigm" for medical practice, EBM aimed to implement the findings of high quality randomised controlled trials (RCTs) and observational studies effectively into clinical practice, policy and guidance. RCTs, which test an intervention within a controlled environment and with a carefully selected population in order to minimise the risk of bias or confounding factors, are highly regarded in terms of validity (200). They are grounded in the positivist paradigm, where a hypothesis can be generated and tested, and a particular truth or reality proposed or disputed, seeking to answer the question "does this intervention work?" Cochrane reviews that collate evidence from RCTs are widely considered the highest standard of evidence for healthcare, and feature at the top of traditional hierarchies of evidence (201).

Ever since the launch of EBM, there has been debate about how the emphasis on experimental evidence may devalue or underestimate the human interactions that occur in the delivery of healthcare to patients (202). In the "real world" of clinical practice, it is rarely possible to control the environment in which a healthcare intervention is delivered. Variables that cannot be controlled include a patient's individual combination of medical conditions, or how likely they are to comply with an intervention such as a drug treatment or medical device. The delivery of EBM at the frontline relies on clinicians to make skilled decisions in partnership with patients and families, carefully applying guidance or protocols to the circumstances of individuals. The willingness and ability of the clinician to do this may be affected by personal factors, such as their previous clinical experiences, the behaviour of colleagues who they consider role models, and wider factors including organisational culture or perceived resource constraints (128).

Palliative care as a complex intervention
Palliative care is an area of clinical practice that is complex, and where the application of positivist EBM is challenging. Broad definitions of palliative care, which describe a philosophy of care rather than a particular service, are one challenge. Other factors influence the delivery and quality of palliative care in practice, such as the availability and accessibility of specialist palliative care services, and timely access to medications, such as morphine to relieve pain (203). Other more subtle and nuanced factors also play a part, including the patient's and family's understanding of their disease and the term palliative care, their values and beliefs, their opinions and their experiences of the healthcare system.

Relative to other areas of healthcare, the evidence-base to inform palliative care practice and service design, particularly for children, is scarce. Ethical and logistical concerns in the conduct of research with patients who may be approaching the end of their lives, and their families, are well documented (12, 204-206). Furthermore, developing evidence-based protocols and algorithms to aid the implementation of policy in such a complex area of practice risks over-simplifying the delivery of care, and failing to capture the patient and family priorities and needs. For example, the documentation of a preferred place of care and death has been widely adopted as a measurable outcome in palliative care commissioning. Research has brought into question the value of documenting a preferred place of care and death, including a systematic review that provided evidence to suggest that many people are never asked about their preferred place of death (207). Another systematic review, which examined preferred place of death for children, identified only nine relevant studies and concluded that the evidence base for current policies stressing the need to increase the numbers of children with life-limiting and life-threatening conditions dying in their preferred place of home is inadequate (114). There is work in progress to identify patient-reported

outcome measures in palliative care (208), including outcome measures for children (209). Until there is a suitable alternative, commissioners who have responsibility for designing contracts with healthcare providers may continue to make use of measures such as preferred place of care and death, despite the limitations of this approach.

4.3. Epistemological perspective

As described in the introduction and literature reviews of this thesis, there is a clear need for further research evidence to inform the future delivery of palliative care to children. A researcher's choice of research method draws upon their personal theories of knowledge and knowing. Their epistemological position underpins how they know or find out about the world, and may depend on their own personal experiences, professional background and training. For example, a clinician with medical training, surrounded by a culture of EBM, may take a positivist approach to research, seeking to identify facts about reality through observation and experiment. A social scientist, based in an academic institution, may take a constructivist approach, which asserts that there is no certain reality, and that all knowledge is constructed through both social and individual phenomena.

I am a medical researcher with a clinical background in general practice. My observations in clinical practice resonate with the problems described around the application of evidence-based guidelines and protocols to individual patients, with their individual healthcare conditions, beliefs, expectations, preferences and social circumstances. In my experience, palliative care is a complex, multi-faceted intervention in which the nuances of human interaction, the changing and unpredictable nature of medical conditions, and the discrete details of every healthcare consultation all play a part. Positivist research does not necessarily lend itself to an intervention as complex as palliative care. Material factors and social realities within the healthcare system exist beyond human consciousness, and are relevant to the delivery of palliative care. This research recognises and acknowledges that these material factors and social realities exist, so research grounded in an entirely constructionist philosophy would not be appropriate.

In order to address the research questions, the epistemological position that I have adopted is one of realism, a position that sits between positivism and constructivism. Realism acknowledges that both the material and the social worlds are real, independent of, and inter-dependent with, human understanding (210). Realism recognises that social systems are complex, open systems that cannot be controlled, and places emphasis on understanding the non-observable processes, or explanations, for *how* and *why* things work. Realism goes beyond what can be measured in the social world, to explain the deeper, causal powers that shape what can be observed (210, 211).

4.4. Methodology

It is rare to see the theory and epistemology that informs research overtly stated in medical research papers, perhaps because of the constraints of word limits or because the researcher has not undertaken their research for a thesis and therefore has not had to consider their position in detail. There is wide variability in the theoretical approaches taken to research in palliative care. In keeping with the emphasis on EBM, but at odds with the broad definition of palliative care provided by the WHO, some published evidence in this field takes a positivist approach, clearly conceptualising palliative care as a discrete intervention or a particular specialist service (212-215). Other studies test a specific aspect of palliative care such as advance care planning (ACP) (216) or medication for symptom control, with some studies being collated into systematic reviews (217, 218). For some aspects of palliative care delivery, this positivist approach is highly appropriate and underpins the effective design of trials to inform elements of practice. However, for broader, more complex issues in palliative care, such as the delivery of palliative care as an approach, other research methodologies may be more appropriate.

This PhD research aims to explore the views of key stakeholders who have experience of healthcare for children with life-limiting or life-threatening conditions, and to gain an understanding of the world from their perspective: family members, healthcare professionals, and the children themselves. The autonomy of the children is prioritised, their views are considered valid and important. Qualitative research is appropriate for developing detailed descriptions and insights of a particular experience or reality, and this research is therefore situated within a qualitative paradigm.

The research design has drawn upon a number of qualitative methodologies. Very little previous research has been conducted with children in palliative care, and elements of this study (recruitment data collection and data analysis) have therefore taken a deliberately iterative, inductive approach, identifying repeated ideas and concepts through the course of the research, informed from the outset by grounded theory. This is a qualitative methodology developed by sociologists Glaser and Strauss in the 1960s which focuses on building theory, taking an inductive approach to data collection and analysis without the use of any existing theoretical framework (219, 220).

Other methodologies were considered as the research plan was developed. Realist methodologies were considered to have distinct advantages, with the explanatory focus of realist research addressing the aim to produce increased understanding into *how* and *why* certain experiences happen for children and their families. Furthermore, a specific aim of realist research is to generate theory to inform policy-relevant recommendations (221).

Realist methodology has been influenced by the work of a number of philosophers. Popper regarded scientific inquiry as a continuous, evolutionary process, and proposed that certainty could not be established through even a series of discrete scientific experiments. Instead, Popper proposed that science and knowledge grow through a cumulating process of theory testing and explanation. This is a fundamental principle of realism (222, 223). Bhaskar subsequently proposed a theory known as critical realism, and the concept of "generative mechanisms"; hidden processes which trigger certain outcomes. He proposed that scientific experiments involve designing, rather than observing, a particular system in which an intervention is tested. Bhaskar proposed that any experiment should be preceded by the development of a theory of how the intervention under study affects the system (224). Following Bhaskar, Merton described the idea of "middle-range theories", which could be formulated to explain the influence of social structures and behaviours in the implementation of interventions or policy. He proposed that the history of an intervention or policy can affect how effective, or not, that intervention or policy is when it is implemented (225). Campbell then described a process known as "theory refinement", based on a need to examine all types of evidence and influence, both quantitative and qualitative, in order to formulate hypotheses that lead to reasoning. These hypotheses can be subjected to repeated testing through the collection of new data which develop and refine the theories further (226).

In 1997, social scientists Pawson and Tilley published their book "Realistic Evaluation", describing an approach to evaluation that is theory-driven and focusses on generative causation. They proposed that reality is generated through a range of causal processes or mechanisms that cannot be seen. The approach they described is interpretive, and designed for the study of complex interventions or "programmes", leading to the development of a programme theory to guide the implementation of policy into practice (85, 86, 187). The approach seeks to bridge the gap between policy and practice by providing an understanding and explanations into how phenomena come about as a result of hidden mechanisms, enacted under certain circumstances; the "what? why? how? who for? and when?" questions about an intervention or "programme" (126, 227). It acknowledges that there are a wide variety of dynamic contexts and mechanisms that can affect whether or not the desired outcome of an intervention is achieved, including geographical and environmental factors, political, social and cultural issues and historical factors.

4.5. Established theories that inform the thesis

Unlike grounded theory, which informed the iterative, reflexive approach to the early stages of the research, realist research often draws upon established theories. The aim and ambition is to develop policy relevant programme theories and recommendations, with those informed by established

theory being more effective than those that are not. Furthermore, formal theories provide relevant insights from other research and knowledge about a topic, which can inform the design of a study, data analysis, and the development of the programme theory (210). Formal theories have informed different stages of this research as follows:

Complexity theory in healthcare
The research conducted for this thesis is grounded firmly in a recognition of complexity in healthcare at every level, from the child and their family, through to healthcare organisations and organisational culture. The research examines and attempts to address levels of complexity related to the healthcare conditions with which children live and the management of these conditions, in the context of a complex healthcare system. Complexity theory dictates that clinical practice, organisation, information, management, research, education and professional development are interdependent and delivered through multiple self-adjusting and interacting systems. The systems are ever changing and dependent upon multiple factors including the behaviours of individuals within them and the availability of resource, and external factors including political agendas and societal culture. These factors bring constant uncertainty and unpredictability to the system (228).

The delivery of healthcare to an increasingly complex population requires a paradigm shift away from traditional "reduce and resolve" approaches to clinical care and service organisation. There is a need to "think outside the box", proposing conceptual frameworks that acknowledge complexity and offer dynamic and creative models for the future delivery of both clinical care and healthcare organisation (229).

Bronfenbrenner's bioecological systems theory
Bioecological systems theory, which describes micro, meso and macro systems affecting human development, has informed this research, providing structure to the data analysis and subsequent development of a programme theory. The micro-system refers to an individual's immediate relationships with other individuals. The meso-system describes the interactions between individuals in the micro-system, and the macro-system describes the wider society and culture in which the individuals live. The biology of the individual was also described as being an influence in the microsystem, hence the term the "bioecological" systems model (230).

Bronfenbrenner first proposed his theory of human development in 1979 (230). Bronfenbrenner was a reflective theorist, and the bioecological systems theory evolved over time. In the 1990s, it evolved such that less emphasis was placed on contextual factors, and more on the individual characteristics of a person. It is an early version of bioecological systems theory that informs this thesis (231).

Theoretical concepts that have informed the thesis.

Other theoretical concepts that have informed the research from other medical disciplines, particularly concepts that relate to the interactions between healthcare professionals and patients. Two theoretical concepts derived from general practice and psychiatry have particularly informed the data analysis. Michael Balint's seminal work, "The doctor, his patient and the illness", describes the consultation as an intervention in its own right. The concepts described by Balint were reflected in the data collection and have informed the development of the programme theory and discussion of findings (232, 233). Similarly, values-based medicine (VBM), a framework developed originally in the domain of mental health, which proposes that the values of individuals are powerful influences in healthcare, clinical practice and research, and that their impact is often underestimated (234), became particularly relevant during the data analysis. Concepts from VBM informed the data analysis, development of the programme theory and recommendations.

4.6. Chapter summary and theoretical framework

This chapter has described some of the existing tensions between EBM and palliative care as a complex intervention, and the philosophies, methodologies and theoretical approach to this research. Figure 4.1 shows a theoretical framework that informs the research. The framework summarises the approaches that have informed the thesis, and links them to the research methods, which are described in more detail in Chapter 5.

Figure 4.1: Theoretical framework for the thesis

Ontology
- Research grounded in clinical practice as a GP (introduction)
- Policy, research evidence and guidance are not delivering the necessary improvements in palliative care

Epistemology
- Palliative care "is" and palliative care "works" - often described as a discrete intervention or specialist service but is more complex than this
- We need research to address the gap between policy and practice - asking the how? why? in what circumstances? questions

Established theories
- Realist theory and complexity theory in healthcare underpin the thesis
- Bioecological systems theory - micro / meso / macro level - structures the analysis and programme theory
- Relevant theorietical concepts from other disciplines: Balint and values-based medicine.

Methodology
- Qualitative
- A deliberately broad and iterative approach to data collection from children and families, derived from grounded theory.
- Realist approach allowing an explanatory focus and the development of a programme theory.

Methods
- Serial qualitative interviews with children and families
- Focus groups with healthcare professionals
- Thematic analysis
- Realist analysis

5. Methods

5.1. Overview of Chapter 5

Chapter 5 outlines the research methods. The research protocol has been published in an open access journal (Appendix 1 (4)). This chapter incorporates and expands on the published protocol to describe the study setting and provide a more detailed explanation of the data collection and analysis methods.

5.2. Study setting

The study setting was the West Midlands, a diverse region of England with densely populated cities and large rural areas. The West Midlands was an appropriate study setting given its highly diverse population in terms of socioeconomic, ethnic and cultural backgrounds. The largest city in the region is Birmingham, which has a population of 1.01 million people. Children represent around 22.8% of the population of Birmingham, a higher proportion than most other UK cities (235). Birmingham Children's Hospital (BCH) is a large children's hospital, providing highly specialist paediatric services to children with cancer and complex cardiac conditions, and those who require liver, small bowel and renal transplantation. The hospital has one of the largest paediatric intensive care units (PICU) in Europe. The first Rare Diseases Centre for children in the world was opened at BCH in 2018. The first consultant in paediatric palliative medicine at the hospital was appointed in 2017.

The population of children with life-limiting and life-threatening conditions in the West Midlands is increasing in terms of both numbers and complexity (236). There is also marked variability in the provision of specialist palliative care services, including for children, across the region, a situation which was highlighted in two reports commissioned by NHS England: West Midlands in 2017 (236, 237).

A report of national and regional prevalence published by Together for Short Lives in 2012 estimated that there were 4,493 children and young people (up to the age of 25 years) with life-limiting and life-threatening conditions in the West Midlands in 2009/2010 (7). The number had risen to 7,704 in the 2017 regional prevalence estimates (236). Other research suggested that over 50% of families with a child known to palliative care services in Birmingham and Solihull were from Black or ethnic minority backgrounds (238), however, this does not necessarily represent the population of children with life-limiting or life-threatening conditions, only those who have accessed palliative care services.

5.3. Data Collection Methods

As outlined in Chapter 4 (Methodology), qualitative research methods were adopted for this research in order to collect detailed, in-depth data to provide insights into experiences of healthcare

from the perspective of children with life-limiting and life-threatening conditions and their families. The literature reviews (Chapter 3) revealed that there is very little research to date that specifically explores children's experiences of healthcare when they have a life-limiting or life-threatening condition, or their views of palliative care. Not only is there a lack of evidence about their views and opinions, there is also very little to describe research approaches that are acceptable and appropriate for the conduct of research with children with life-limiting and life-threatening conditions (239). The data collection methods therefore developed iteratively and with the intention that they were reflexive according to the needs of the children and their families. The concurrent processes of data collection and data analysis outlined in the grounded theory approach informed this inductive approach. This approach allowed reflection and careful consideration of the initial themes and codes generated through the course of the interviews. It also allowed for reflection about what methods and interview techniques worked effectively during the interviews with children, what worked less well, and any emerging themes it would be helpful to explore in further depth with children and families in subsequent interviews or interviews with other children and families (220, 240, 241).

Data collection took place in two overlapping phases: (1) serial child and family interviews, and (2) focus groups with healthcare professionals involved in the delivery of paediatric palliative care. Both semi-structured interviews and focus groups are suitable methods for complex, emotionally charged subject areas (242, 243) and had significant benefits over other methods for the purposes of this research. Both allowed for a flexible approach, with active listening, reflection and appropriate adaptation of each interview and focus group depending on the circumstances of participants and the issues raised.

5.4. Serial child and family interviews

Serial qualitative interviews were the data collection method of choice for this research with children with life-limiting and life-threatening conditions, whose developmental, clinical, psychological and emotional needs are individual and can fluctuate rapidly. Interviews would also allow for subtle and nuanced aspects of communication to be observed and captured that would be lost through other research methods such as questionnaires, written surveys and case note reviews. Conducting serial interviews provided benefits including building rapport with children and their family members, and being able to observe the changing needs of the children and their families, and their experiences of healthcare services, over time (244). As the children became more familiar with me over the course of the interviews, they became more willing to take part in the interview conversations. A further benefit of serial interviews was that the interview process could be tailor made to the needs of each child and family. Family members chose the time intervals in between

the interviews and the location for each interview, depending on what was convenient for them and their personal preferences.

A major consideration in conducting research in palliative care is effective recruitment. Despite evidence to suggest that taking part in research can provide benefits for participants (245), there are many persistent barriers to recruitment (12, 246). Potential barriers to recruitment in this study included the unpredictable nature of the child's condition and the impact of medical treatments and technology on the child's ability to consent and take part in research. Gaining access to children and families through healthcare professionals, who may have their own ideas and concerns about the child and family and the research process, can also be a barrier to recruitment (known as "gatekeeping") (111, 247).

The approach to recruitment for this study was planned carefully, taking into account the factors that could affect a child or family's ability to take part including the clinical condition of the child, conflicting demands on the family's time, their own motivation and understanding of research, and accessibility of the study through their clinical teams.

There were two approaches to participants:

1. By direct invitation via their clinical team
2. Via leaflets and posters displayed in public areas in the hospital (such as notice boards on wards and in outpatients).

Recruitment via Clinical Teams

The research plan was introduced to clinical teams in both the hospital and the community through formal presentations and through meetings with individual clinicians. The research was also presented to the regional paediatric palliative care network. A "snowball" approach was taken to meeting with clinical teams and clinicians, with interested clinicians putting me in touch with their colleagues and inviting me to relevant meetings. The result of this approach was that formal and informal meetings were held at BCH with the clinical teams who expressed an interest, including the hospital complex care team, paediatric intensive care staff, the palliative care team, the liver team, cardiac surgeons, neurosurgeons and paediatric surgeons. I undertook periods of shadowing with clinical teams, on hospital wards and in outpatient clinics including with the hospital palliative care team, the acute pain team, oncology and renal team. An honorary contract was in place in order to allow me to undertake these periods of shadowing. These experiences provided not only access to potential participants but also the opportunity to observe and learn about different clinical environments, which was a valuable experience in my joint capacity as both researcher and GP. I

also gave presentations about the research plan at the Birmingham Community Healthcare Foundation Trust community paediatricians meeting and the West Midlands Paediatric Palliative Care Network meeting, and provided information about the research to the children's community nursing and palliative care teams.

Any children and families who were identified as potential participants were provided with a participant information sheet. There were separate information sheets for young children, older children and families, which had been designed with the PPI group (Appendix 5). The information sheets contained detailed information about the project and how to be involved, as well as my contact details. Potential participants expressed their interest either directly to me, or to a member of their clinical team. They provided a preference of phone, text message or email, for me to make contact to discuss participation in the study. If the child and family were willing to participate following this discussion, a date and time for interview was arranged.

Recruitment via Posters
Posters were displayed in outpatient waiting rooms at the hospital. The posters provided details of how to contact me directly to express an interest in participating in the study.

Inclusion and Exclusion Criteria
The inclusion and exclusion criteria underwent several revisions before being finalised. Concerns about the term "palliative care" were raised during PPI work, where group members stated that they disliked the term and requested that it be considered more carefully in the study. Following this feedback, careful consideration was given to the inclusion and exclusion criteria, and participant information sheets. A decision was made to ensure that participation in the study did not depend on the child receiving care from a specialist paediatric palliative care team. This allowed for inclusion of any child with a life-limiting or life-threatening condition, regardless of whether or not they were known to palliative care services. Further thought around the definition of life-limiting or life-threatening condition for the purpose of the study was then required. Children who had a life-limiting or life-threatening condition, as defined by Together for Short Lives (2), and who had had a life-threatening episode, resulting in admission to the paediatric intensive care unit (PICU), were included, as well as those with relapsing or remitting disease. The rationale behind broadening the inclusion criteria in this way was that it would potentially allow investigation into views of the term "palliative care" amongst children and families who were not known to specialist services, and provide insights into some of the facilitators and barriers to referral to specialist paediatric palliative care teams. This approach raised a number of ethical concerns, which required careful consideration, as outlined later in this chapter.

The age range for the study was 5-18 years (school age children). Neonates, pre-school children and young people aged over the age of 18 years were excluded. Specific issues around healthcare services arise when considering neonatal care and young people who are making the transition from paediatric to adult services, both of which warrant research in their own right. Data collection methods and interview techniques would need to be designed and tailored to the needs of pre-school children; this is also an area for potential future research.

The final study population comprised children with life-limiting or life-threatening conditions, aged from 5-18 years, and their family members, some of whom had experience of a palliative care service, and some who did not. Table 5.1 provides the inclusion and exclusion criteria:

Table 5.1: Inclusion and Exclusion Criteria for child and family interviews

Inclusion Criteria	1. Children aged 5-18 years (school age) with a life-limiting or life-threatening condition who are under the care of the Community Children's Nursing Team and/or the Children's Hospital and who either: - receive palliative care services - are aware of (have had discussions about) palliative care services - are living with relapsing or refractory disease - or have had a life-threatening episode (admission to the Paediatric Intensive Care Unit (PICU)) 2. Their family members, who live in the same household.
Exclusion Criteria	- Children aged < 5 years and > 18 years. - Families of children < 5 years and > 18 years old - Children and families with whom I have clinical contact - Children and / or families who do not wish to participate. - Children who are too unwell will not be approached for interview, but their family members may still participate if they wish to. - Children who are unable to participate in a conversational interview for any reason related to their condition will not be approached for interview, but their family members may participate if they wish to. - Children and families who are unable to provide informed consent in English will not be approached for interview.

Interview procedure

The aim of the study was to capture the views of children and their family members, and to provide insights and understanding into their experiences of the healthcare that they receive. Each interview took into account the needs of the child and family, and the child's capabilities, depending on whether they had any kind of learning disability or communication difficulty associated with their condition, and their state of health on the day of the interview. This included the consent and agreement processes. Parental consent was obtained for every interview. Children were provided with the opportunity to sign an agreement form, but did so only if they chose to.

Interviews were carried out in a range of locations including the children's homes, inpatient wards and outpatient clinics, according to the preference of participants. Children and their families were asked to express a preference for who would be present, and interviews were conducted either with individuals or with the child and family member together.

Interviews were deliberately open and conversational, using a blended approach of interview techniques. An initial topic guide provided some structure for the interview, but this was not prescriptive and was not used in the same way that a questionnaire would be (Table 5.2).

Table 5.2: Initial topic Guide for child and family interviews

For all families	For those aware of "palliative care"
Introduction **Please tell me your story, in any way that you can / want to** **Please tell me the story of you** *Your Story* **Please can you tell me about you?** **Your family?** **Your child(ren)** **What is important to you?** **What do you like to do?** **Which places are important to you?** **Where do you spend your time?** **Which services are involved in your care?** **Who comes to see you?** **What do they do?** What's helpful? What's not? **Which healthcare professionals do you consider to be key in the delivery of your care?** **What works best?** Which services / professionals are most helpful? Which services / professionals do you value most? **What works well? What doesn't work?** **How do you think services could be improved?** **Do you talk to other children / young people / families about your healthcare / services"?** What do you tell your friends? What tends to come up in these discussions? Would you recommend these services to others?	*Palliative care and you (if appriopriate)* **Do you have "palliative care" services?** Have you ever heard the term "palliative care"? What does that mean to you? What do you receive those services for? What do these services provide for you? Does it matter what a service is called? Do you receive services from the hospice? **Can you tell me how you came to receive palliative care / know the palliative care nursing team / the hospice?** When were you referred? Who brought it up / made the referral? How was this discussed with you? How was that for you / your family? **Do you think that medical / nursing staff receive enough training in this area?** What makes you think that? **Anything else?**

Rapport building was particularly important from the outset of each interview, and throughout the interview process, both for the children and their family members. Establishing rapport with the children was a gradual process, and depended on a number of factors, including whether or not the children had met me before the interview (for example when I was shadowing their clinical teams), or during a previous interview. As they became more familiar with me, they tended to offer more during the interviews. Building rapport with the families, and developing an appreciation of the daily challenges they faced, was a key element to enabling subsequent interviews or understanding why these were not possible and making a decision to stop contacting the family if necessary. In this situation, I provided them with an open invitation to contact me again for a further interview if they wished to.

The structure of the interviews developed iteratively, but each interview was open and conversational. Passive interviewing techniques, including the use of open questions, which allowed the participant space and time to tell their story, were employed. The first interview with each child and family began with "Please tell me your story, in any way that you can". Advanced communication skills such as active listening, responding to verbal and non-verbal cues, summarising, reflecting back and using silence were all important to ensure that the children had the space and time during the interview to participate as much as possible, to provide assurance that they had been heard, and to check understanding (248). Active interview techniques were used to explore specific aspects of care that were raised by children and families, including questions which took an appreciative inquiry approach, asking "what works well?" and "why does it work well?" (249, 250).

Field notes were made following every interview. These included notes about interview techniques and strategies, and adaptations to the topic guide made according to the participant's responses. An ongoing process of reflection and note-making provided opportunity to rehearse questions and techniques ahead of interviews with children and families if particular issues were to be explored. Areas that were explored during subsequent interviews related to the "how?" and "why?" certain events or experiences happened for children and families, from their perspective.

For interviews with children, a range of techniques were employed including de-personalising questions, developing a narrative in the third person, and using props and toys to encourage story-telling. Arts-based activities including drawing, stickers and bracelet making were used with children either as a focus of the interview to facilitate questions (using techniques including draw-write-tell) or as a mutual activity alongside which the interview took place (239, 251-253). Often these were

activities that the child had expressed a preference for during a previous interview. Some of the props and activities are illustrated in photographs 5-8:

Photograph 5: Bracelets made by a child (aged 17 years) during an interview as gifts for healthcare professionals

Photograph 6: Key rings made during an interview with a child (aged 17 years)

Photograph 7: Rabbit puppet made during an interview with a child (aged 6 years)

Photograph 8: Toy healthcare professionals arranged into families by a child (aged 5 years) during an interview

Photograph 9: Important characteristics of healthcare professionals from the perspective of children
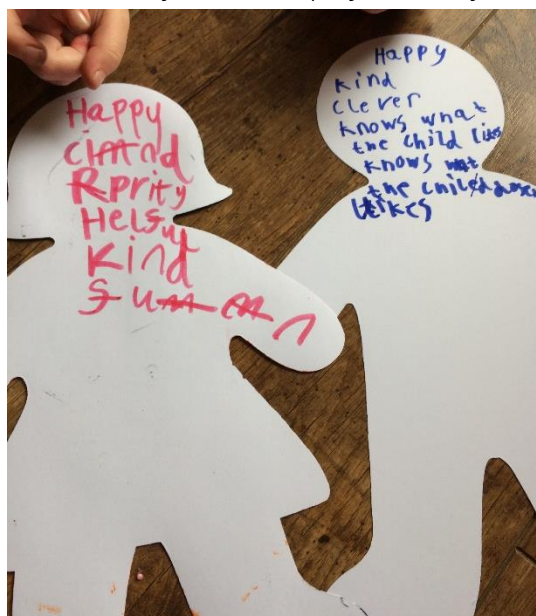
PPI advice informed the format of interviews for children (Table 5.3), with feedback and suggestions from PPI group members incorporated into the interview plan.

Table 5.3: Feedback from PPI groups that informed the interview plans

January 2016	*"Those who are passionate about improving palliative care will take part regardless of how sensitive this may be"*
July 2016	*"Remember young people who are seriously ill are more mature, they have to grow up"* *"Keep it simple as often a child will openly speak anyway"* *"'Do you talk about it to your friends?' is a good question, a good way to talk to most ages."* *"Children are more eloquent, mature and more capable than you think"*
October 2016	*"Use pictures and images, more emojis"*
February 2017	*"Doesn't make me uncomfortable as I think it is very important and relevant"*

Every interview was audio-recorded. The field notes made during and after the interview included any observations made about what was happening for the child and family, their family structure, important events and any strong themes that arose during the interview that would warrant exploration at a subsequent interview. Field notes included detailed reflections of the interview process, including elements that had gone well or less well, to guide future interviews, and potential questions for future interviews. Observations of the family situation included (with family permission) some photographs, which illustrated issues that they had described during the

interview, for example a bedroom completely dedicated to notes about the child's care, or the adaptations made to the house, or a shed built in the garden in order to accommodate a stock of equipment.

Field notes were also made to record any other contact with families in between interviews. Family members chose whether to keep in touch in between interviews and if so, how they would prefer to do so. Chosen methods were text messages and email. Two families invited me to follow their blogs on social media, which provided further insights into their experiences and helped with timing future contact and interviews because they chose this forum to document significant deteriorations in their child's condition, and hospital admissions. Telephone conversations to arrange interviews frequently resulted in long conversations about what was happening for the family. On several occasions there was a chance meeting with a family while I was at the hospital for another reason; bumping into a family who were attending an appointment in clinic at the same time that I was going to conduct an interview with another family, for example.

Occasionally interviews had to be postponed due to a deterioration in the child's clinical condition, or were interrupted due to a conflicting demand on the family's time (often clinical). On one occasion, the family were not home when I arrived to conduct the interview due to a sudden deterioration in the child's condition that had resulted in an unscheduled visit to hospital. On another, clinical staff arrived to discuss updating an ACP. During interviews in the hospital, clinical staff frequently visited the children and their families, or the children had to leave the room for treatments. Clinical environments such as isolation rooms required particularly careful consideration when interviews were to be conducted there. No interviews were conducted in oncology isolation rooms. One interview was conducted in a side room with a young person who was isolated due to the possibility that she had infectious diarrhoea. Ward barrier nursing procedures were followed, participant information leaflets were presented in plastic wallets that could be wiped with sterilising wipes, and the audio-recorder sterilised following the interview.

Recruitment to serial interviews
Issues that affected recruitment of families to subsequent serial interviews were frequent. One interview was halted as the child was experiencing significant pain. Another was deferred because the family had had to take their child to hospital suddenly and were not at home, and another was conducted over two dates after the palliative care team arrived at the house during an interview. My clinical background helped me to gauge an appropriate time to stop and rearrange the interviews when necessary, and to ensure that the family prioritised clinical activities, such as a nurse's visit, over the interview process. One family almost turned the nurses away so that they could continue

with the interview. They required reassurance that it was more appropriate to see the nurses, and that I would return to complete the interview another day.

Decisions about how and when to make contact to arrange an interview with a family following bereavement were difficult. An important aim of each interview was to ascertain the expectations of the children and their family members regarding the child's condition and what might happen in the near future. The family chose a timeframe for the next contact following each interview. Methods of contact were also the choice of the family – phone, text message or email. No specific plans were written into the study protocol about how or when to make contact with families in the event of the death of the child; this provided a dilemma about contacting the family again and should not be omitted from future studies. In this study, this situation arose with one family. Contact was made during the timeframe and by the method previously agreed with the family (text message). They declined the interview at that time, and were offered the opportunity to be interviewed later on if they wished to, but they were not repeatedly approached. Hospital bereavement and family liaison teams have processes and procedures in place for making contact with bereaved families; these are valuable resources to inform plans for follow-up during bereavement in future studies.

5.5. Healthcare professional focus groups

There are 12 paediatric palliative care networks in the UK, which include professionals from a range of organisations within paediatric palliative care. The networks provided accessible forums in which to conduct focus groups that would capture the views of a range of professionals, at a time when these professionals were already meeting together.

Recruitment

A pragmatic approach to recruitment to the focus groups was taken. The research proposal was publicised and presented at regional and national conferences, and the networks invited to take part in the focus groups via the network chair-people. Following these presentations, four network chair-people expressed an interest in their network's participation in the study. Arrangements were made to attend their pre-arranged network meetings in order to run a focus group.

Participant information leaflets were emailed to all network members prior to the meeting. Paper copies were provided at the meeting (Appendix 6). Consent forms were completed at the time of the focus group. Participants were reminded that they were under no obligation to take part if they did not wish to, and given the option to provide written comments using a sheet designed for this purpose if they preferred.

Inclusion and exclusion criteria

Focus group participants were all members of a regional paediatric palliative care network, and all members were invited to participate. The professionals who took part in the focus groups were all involved in the delivery of palliative care to children in some way. Not all had been through specialist training programmes in either medicine or nursing, but many had years of experience in the field. There were also allied healthcare professionals, in whose profession there was no specialist training in palliative care. All had a particular interest in palliative care for children.

Focus group procedure

The focus groups were carried out at existing paediatric palliative care network meetings in order to minimise inconvenience to participants. A presentation of the emerging study findings was followed by a structured focus group discussion that aimed to test out the emerging themes from the analysis of the child and family interviews, and to refine and refute the developing context-mechanism-outcome configurations as the study progressed by collecting data about the views, experiences and perceptions of professionals. The focus groups were all audio-recorded. A topic guide provided structure to the focus group discussion (Table 5.4). The topic guide was deliberately broad at first and developed iteratively for each focus group.

Table 5.4: Topic Guide for Focus Groups

Question	Prompts
What do you consider to be the most important elements of "palliative care"?	Who is involved? What are professionals having to do to make things happen for families? How are they Introduced to children and families? Are language / understanding barriers? • Are findings from the interviews as expected? • If not, what is surprising?
How is palliative care delivered at the frontline, in your experience?	What does it take? What are the experiences / actions / views of individuals in order to ensure palliative care is provided to children and families Are there any examples of good practice? And less good? What are the facilitators and barriers? What are views on access to specialist palliative care? What is specialist palliative care and who should provide it?

	What are views on the organisation of services? How could this be improved?

5.6. Data Analysis Methods

Recordings of interviews and focus groups were transcribed verbatim, and field notes were transcribed into Word documents. As the researcher, I transcribed eight of the child and family interviews, with the others outsourced to a university-approved professional transcription service. The transcription service manager was briefed about the nature and content of the interviews, and support offered if necessary. I transcribed all of the focus group recordings. The benefits of undertaking this transcription for me as the researcher included the opportunity to familiarise and immerse myself in the data, reflecting on what was said, who by, and how it was said.

The transcripts were fully anonymised with personal identifiers removed and each participant assigned a unique identifier code. Names and places were also anonymised using [name of friend], [name of nurse], [hospital], [city]. Interview data that could identify the child, families or professionals involved in their care because of the individuality and context of the narrative were included in the data analysis, but excluded from reporting. The transcripts were uploaded into the qualitative data software package NVivo11 for data handling.

Thematic Analysis

Data analysis began alongside data collection with a broad thematic analysis. The child and family interview transcripts were analysed first, with the initial findings informing the focus group presentations and topic guide.

It was anticipated that that the longitudinal data collected from the child and family interviews would lend itself to longitudinal data analysis, to describe the changing experiences of the child and family over time (244). This might have involved developing innovative approaches to the data analysis, as in previous palliative care studies, such as the use of matrices to compare and contrast themes arising from interviews at key times for families, including the time of diagnosis, an admission to intensive care or referral to a palliative care team (244). However, as the interviews and analysis progressed, and the richness of the data became clear, a decision was made to continue, strengthen and formalise the thematic analysis using an inductive approach as described by Braun & Clarke (81, 219), in order to capitalise on the richness of the data that had been gathered.

The analysis began alongside data collection with a process of familiarisation, reading and re-reading the transcripts. A continual process of reflection and note-taking, using techniques including the One

Sheet of Paper (OSOP) technique to bring together the themes that were emerging from the data (254) accompanied familiarisation with the data. Following familiarisation, a descriptive code was applied to every item of data. The codes were developed iteratively; they emerged from the data rather than through the application of a framework. The codes were gradually and iteratively grouped into broad overarching conceptual categories, with the emerging codes and concepts being discussed with the supervisory team (JD, A-MS and JC) at regular intervals (at least monthly) throughout the data analysis process. This allowed for the development of the themes and decreased lone researcher bias (255).

The focus group transcripts were analysed after the child and family interviews, using a framework developed from the main themes from the child and family interviews to guide the initial analysis. As the analysis progressed, data from the focus groups was triangulated with data from the child and family interviews, and new codes and themes that emerged supplemented the findings of the child and family interview analysis. During writing up, healthcare professional views were incorporated to illustrate and provide a broader perspective into the experiences and perceptions of children and their family members.

Applying realist logic to the data analysis
At the time of publication of the study protocol, an in-depth narrative analysis, using structure-form analysis to examine not just what was being said, but how it was being said, was proposed (4, 213). The aim was to undertake this analysis to position the narratives in the context of palliative care delivery from a micro (immediate clinical team), meso (local organisation) and macro (wider healthcare system) level perspective (256, 257).

After careful consideration, and further relevant training, the plan for this phase of the data analysis was changed, and a realist method of data analysis, as explicated by Pawson and Tilley (126, 258) was considered preferable to narrative analysis techniques. Application of a realist logic to the findings of the thematic analysis provided opportunity to focus on generative causation, to interpret and explain the "hidden" mechanisms, triggered in certain contexts, to enable the delivery of palliative care to children and families. The realist approach also provided the benefit of being theory-driven, aiming to generate new theories to lead on to the proposal of policy-relevant recommendations.

Rationale for the application of a realist logic to the data analysis
Two main approaches in realist scholarship have gained prominence since Pawson and Tilley published Realistic Evaluation in 1997 (126). Realist evaluation is primary research, involving the collection of new data from original sources. Realist review is secondary research involving an

iterative systematic search for existing data, followed by analysis, synthesis and interpretation informed by a realist logic. In both realist review and evaluation, context-mechanism-outcome configurations (CMOCs) are developed. Contexts and outcomes are described, and a process of abstraction leads to the proposal of mechanisms, that are triggered in certain contexts to produce the outcomes. Mechanisms are real, but they are not visible; they are triggered in particular contexts to produce outcomes. This study was considered a realist inquiry, comprising a realist literature review and investigation into a broad, complex intervention, rather than an evaluation of the implementation of a more defined intervention.

Application of a realist logic to the thematic analysis resulted in the development of a series of CMOCs. Subsequently, by bringing together the CMOCs, a programme theory was developed to describe what the intervention or "programme" of palliative care comprises, and the sequence of events that must take place in order for a particular outcome to be achieved. The resulting programme theory informed policy-relevant recommendations.

Methods: Realist analysis of the empirical research findings
A realist logic was applied to the findings of the thematic analysis. The coded dataset was interrogated for sections of text that could be used to inform CMOCs. As explicated by Pawson and Tilley, a context was defined as a pre-existing structure, setting, environment, circumstance or condition that influenced whether or not certain behavioural and emotional responses (i.e. mechanisms) were triggered. An outcome was the impact of mechanisms, triggered in certain contexts, and a mechanism was the often hidden, behavioural or emotional response triggered in a certain context. Sections of text were examined to ascertain whether they were functioning as a context or outcome, and the hidden mechanisms described through a process of abstraction. The supervisory team (JD, JC and A-MS) and I regularly debated, compared and consolidated potential CMOCs. Data from the focus groups provided further insights to refine or refute the emerging CMOCs. The CMOCs described the causal relationships between context, mechanisms and outcomes; they proposed how outcomes may be achieved through mechanisms being triggered in certain contexts (227).

The following questions were developed as a guide to this process:
1. What does the data tell us about the important factors in relation to palliative care for the child and family?
2. Is the section of text referring to context, mechanism or outcome?
3. What is the context? What outcomes are described? What are the hidden mechanisms?
4. What is the CMOC?

5. Is the data trustworthy and rigorous?

Methods: Development of a programme theory

The overall aim of the realist analysis was to formulate a refined and testable, policy relevant programme theory, where the "programme" was the delivery of palliative care to children and their families. The overall programme theory brought together the findings of the literature reviews and the empirical research through the following steps:

1. Identification of an initial programme theory that palliative care for children "works" from policy statements and the systematic review.

2. Testing and further development of the programme theory through a realist literature review to identify the CMOCs that provide insights into how palliative care for children "works", and in what circumstances

3. A process of refining and refuting the CMOCs from the realist review with findings and CMOCs from the empirical research, to understand how, when, in what circumstances and why palliative care "works" for children and their families.

The following questions were devised to guide the process (130):

1. What does the data describe about the important factors in relation to the experience of palliative care for the child and family?

2. Is the data referring to context or outcome?

3. What are the mechanisms triggered in this context to produce the outcome?

4. What new insights are provided? Does this evidence refine or refute the corresponding CMOC from the realist review?

A multitude of potential CMOCs exist in this field, so none of the CMOCs or the diagrams that illustrate them in Chapters 9 and 10 are intended to be definitive or exhaustive. The CMOCs have all been extrapolated and proposed using the data yielded from the empirical research, and have been prioritised based on whether they have the potential to refine or refute the programme theory developed through the realist literature review. The overall programme theory provided the basis for policy-relevant recommendations, which have been presented in the Discussion chapter (Chapter 11), along with consideration of the implications for policy and practice.

5.7. Ethical considerations and approvals

Ethical approval was granted in September 2016 by the NHS Health Research Authority (IRAS ID: 196816, REC reference: 16/WM/0272).

Research with children raises ethical and legal considerations around recruitment, consent, and data collection (259, 260). In addition, research regarding palliative care can be emotionally demanding and distressing for those involved. There were also particular ethical issues to consider given the longitudinal nature of the study (261). The children and families recruited to the study were potentially vulnerable, with a risk that they were experiencing considerable distress related to their situation. This had to be balanced with the justification for the research, which was that children and their families in this situation are rarely asked about their experiences. Conducting research to understand their experiences is essential in order to be able to design and develop services that respond to their actual needs.

The ethical issues raised by this study are summarised here:

Language
Published literature suggests that the term "palliative care" is poorly understood and perceived negatively (16-18), a view confirmed during PPI work for the study. A decision was made to ensure that the scope of the study was to investigate the experiences of healthcare of children with "life-limiting", "life-threatening" and "conditions which may or may not get better", whether or not they received care from specialist paediatric palliative care services. The term "palliative care" was not included in participant information sheets. This decision reflected and respected the views those who had taken part in the PPI to advise the study. During the interviews, the term "palliative care" was discussed only if the children or family members raised it, or if there was a verbal or non-verbal cue to start a conversation about it. They were empowered to discuss their views of healthcare, whether or not this specifically included "palliative care".

Recruitment
Recruiting via clinical teams raised a risk of inadvertent coercion to the study by clinicians who knew the family well. In order to facilitate the autonomy of potential participants, clinicians only provided the study information to children and families, rather than actively recruiting them; the initial expression of interest came from the family to me as the researcher. The study plan was discussed with them either in person or by phone and any questions answered before arranging a time for interview. Participants were made aware and regularly reminded that they could decline to take part or to withdraw from the study at any stage without having to give a reason. Interviews were only carried out at a time that was mutually agreed and minimised any potential inconvenience or intrusion for the child and family.

Recruitment through clinical teams is widely used in palliative care research but may be limited by "gatekeeping" (111), which brings a risk that children and families who do wish to participate in research do not find out about these opportunities through their clinical team. In order to address this, posters and leaflets (which were designed in partnership with the PPI group) were displayed on hospital wards and in outpatient clinics. These included my direct contact details as the researcher (email, text and phone), allowing families to express their interest in participating independent of their clinical team.

Within the time and resource constraints of the study, interviews were conducted only with children and family members who could provide informed consent and take part in the interview in English.

Consent

The study raised ethical and legal issues related to consent procedures for children who were under the age of 16 years or who did not have the capacity to consent. Consent procedures were designed with the aim of obtaining written and / or verbal consent and agreement from every individual for every interview. For children under the age of 16, written consent was obtained from the parent and then verbal or written agreement obtained from the child. An agreement to participate form was completed by the child if they chose to do so, in order to respect their autonomy in so far as was possible.

In keeping with the Mental Capacity Act (MCA), there was an assumption of capacity in young people aged 16 years and over, so they were asked for consent first, followed by agreement from their parent(s). Their parental agreement was not legally required, but conducting an interview with a young person about a potentially difficult subject without the knowledge or agreement of their parents would raise further ethical concerns. Where there was concern that the child lacked capacity or was particularly vulnerable, for example with a learning disability, parents were asked to provide verbal and written consent in addition to the child's agreement. Parental consent was required for all interviews carried out in the family home (260).

For a child on a full care order, social worker consent would have replaced that of parental consent, and where possible parental consent / agreement would also have been sought. This situation was not encountered during the data collection.

Interviews

The interview plan was designed carefully to ensure that the risks and burdens associated with taking part were minimal. This was particularly relevant as recruitment to the study may have

occurred soon after sensitive conversations, and the subject areas discussed during interviews had the potential to cause distress to participants.

The interviews were deliberately informal and reflexive to accommodate the needs of the participants. The interview was halted if any of the participants experienced difficulties, such as tiredness or distress. Information about local services and resources for support were available if necessary. Every interview was arranged at a time and in a location of the participant's choice. If this was in hospital, I liaised closely with clinical teams so that the research did not interfere with routine clinical care and ward work.

Minimising potential burdens from serial interviews
At each interview, the family views and understanding of what might happen next as a result of the condition of the child were gently explored and discussed sensitively. When it seemed likely that there would be a deterioration in the child's health, the child and family expectations were ascertained and an agreement made about whether they wished to continue to participate in the study. This was checked again before each interview through contact with the family via phone, text or email, depending on their preference.

Minimising harm to the researcher
The researcher – participant relationship presented possible ethical concerns due to the longitudinal nature of the study, where rapport building was an integral part of the interview process. It was made clear throughout that as a researcher, it was not my role to provide personal support or clinical advice. There were times when these boundaries needed to be reiterated particularly because of my clinical background as a GP. If clinical concerns were raised by the family, we would ensure that there was a clear plan for them to raise these with their usual clinical team.

The nature of the study also raised the risk of emotional distress for me as the researcher. This became increasingly relevant during the processes of immersion in the data and data analysis, rather than during the data collection. Having an experienced supervisory team who also had clinical backgrounds was an important source of support. Formal counselling support was also accessed during the data analysis process via the University of Warwick University Counselling Service.

Serious concerns and safeguarding
Reporting procedures were in place in case any situation was identified that indicated a serious clinical or safeguarding concern or any issue that jeopardised the safety of the participant or another person. No such issues arose during the data collection.

5.8. Chapter Summary

This chapter has provided details of the research methods and procedures that were undertaken in order to conduct the empirical research, underpinned and informed by realist methodology. The data collection methods were qualitative in order to gather in-depth, detailed data about the experiences and perceptions of children with life-limiting and life-threatening conditions and their families. The data analysis processes, thematic analysis followed by the application of realist logic, were designed to lead to the development of a programme theory that could result in policy relevant recommendations. The methodological strengths and limitations are discussed later in the discussion chapter of the thesis (Chapter 11).

Part Three: Findings

The findings of the empirical research are presented in part three of the thesis.

Chapter 6 begins with a report of the outcomes of the recruitment strategy and an introduction to the study population. The children and families who took part in the study are described first, followed by a description of the professionals who took part in the focus groups. Chapter 7 provides the findings of the thematic analysis that relate to the child and family experience of life with a life-limiting or life-threatening condition and interactions with healthcare at an interpersonal (micro-system) level. Chapter 8 provides the findings of the thematic analysis that describe the interactions of the child and family with the healthcare system at an organisational (meso and macro-system) level. A realist logic of analysis is applied to the findings of the thematic analysis, drawing together and describing the contexts, mechanisms and outcomes from the thematic analysis to propose explanatory CMOCs in Chapter 9.

6. Outcomes of the recruitment strategy

6.1. Overview of Chapter 6

This is the first of four findings chapters. The chapter outlines the recruitment of participants, with an introduction to the children and families who participated in the interview study, and the participants of the focus groups. As outlined in the methods chapter, recruitment of children, and people with life-limiting and life-threatening conditions to research presents significant logistical and ethical challenges. Recruitment strategies and experiences are under-reported in the published literature; hence, the outcomes of the recruitment strategy are presented in detail in this chapter, with the intention that this will form the basis of a future publication.

6.2. Recruitment and study participants

Recruitment: Children and families

Recruitment began in October 2016 following the issue of Health Research Authority (HRA) and NHS trust approvals. The initial intention was to purposively sample 12 children, with the aim to recruit three children from each of the four Together for Short Lives categories (2). Following feedback at the PhD upgrade panel in 2016, the recruitment target was increased to 14, to allow for attrition from the study. The aim to recruit three from each of the Together for Short Lives categories was also modified to recruitment of children with a diverse range of life-limiting and life-threatening conditions. 14 families were recruited between October 2016 and June 2017 (4). The first 14 families who expressed an interest in participation and provided consent were included in the study. No children or families were turned away from the study based on their circumstances or conditions,

which, given the nature of the study, was an appropriate approach. Of these 14 children, 13 were recruited via clinical teams, and one family was recruited after responding to a poster in outpatients.

Six other families were invited to participate by their clinical teams, and expressed an interest in the study. After provision of study information they did not consent to participate. The families were not asked to provide a reason for not wishing to participate, and three did not. The other three chose not to participate for a variety of reasons. For one of the families, their child became critically unwell and died soon afterwards. Another family was expecting a new baby and did not want to commit to the study. Another family were already involved in a research study and did not wish to participate in another.

The routes to recruitment via clinical teams are outlined in figure 6.1 below. As described in Chapter 5, section 5.4, the study was presented to as many clinicians and clinical teams as possible (Figure 6.1, column 1). Recruitment then depended on actions of individual clinicians, who invited families to participate (Figure 6.1, column 2). The reasons for the interest and motivation to recruit families to the study were not explored, but would be worthy of further investigation in future studies. The numbers of children and families recruited via each team are detailed in Figure 6.1, column 3.

Figure 6.1 Recruitment strategy
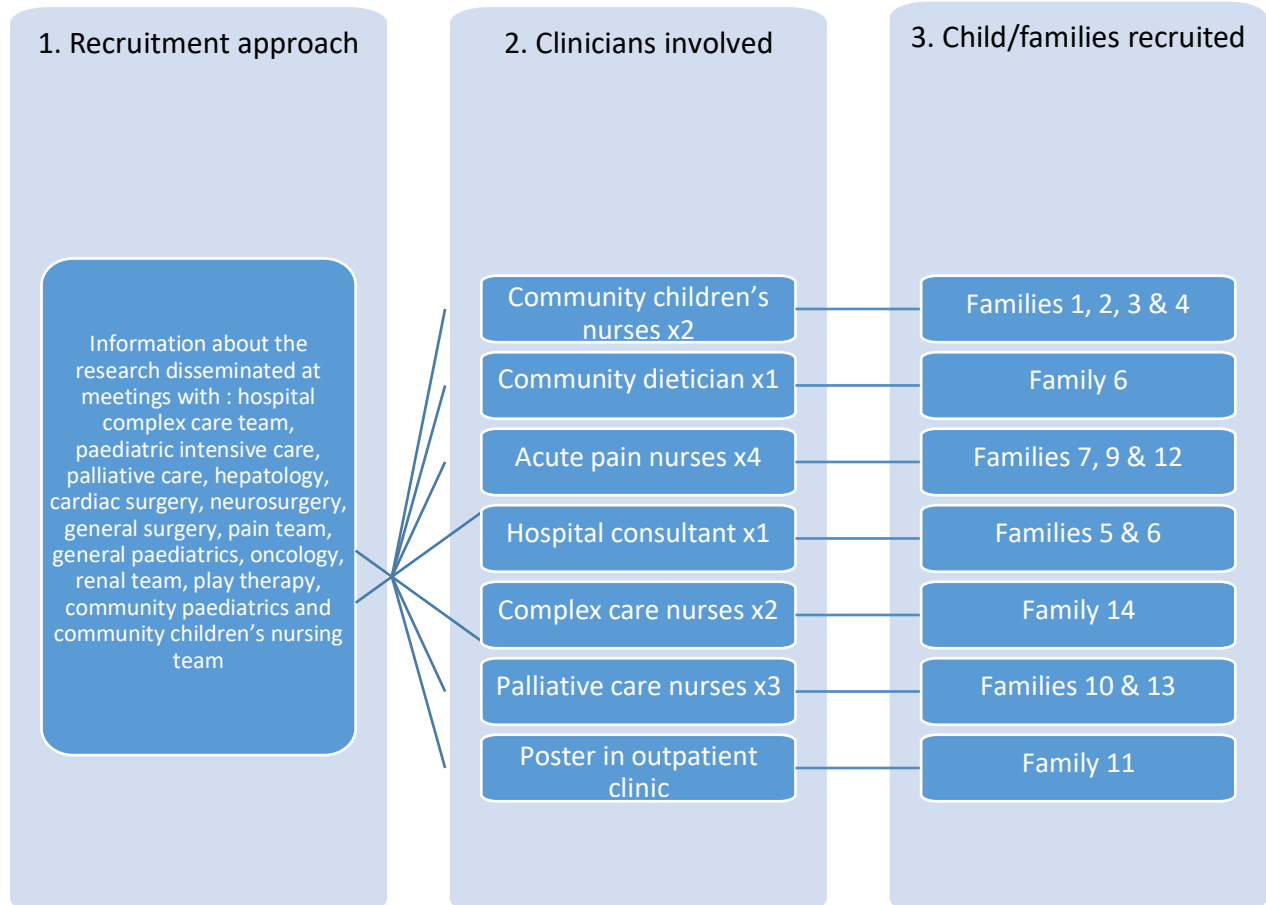
Study population: Children and families

41 interviews were carried out with a total of 31 participants over 13 months from October 2016 to November 2017. Ten of the participants were children with life-limiting or life-threatening conditions, 13 were mothers, six were fathers and two were brothers. One brother (B008) was eight years old at the time of recruitment, the other brother (B002) was 22 years old. The two brothers requested to participate following the first interview with their sibling and family members, and each participated in one of the following serial interviews. Ten of the families were White British, three were Asian and one was African. Of the 13 mothers who took part in the study, two were in full-time employment, four were in part-time employment, one was self-employed and the other six were full-time carers for their children. Two of the fathers who took part were in full-time employment, two were self-employed and the other two were full-time carers for their children. Other fathers, who were either in full-time employment or were self-employed, chose not to participate.

Nine of the children with life-limiting or life-threatening conditions were boys and five were girls. They ranged in age from five to 18 years, at the time of recruitment. The median age was nine years. Three could not participate in the interviews because they had little or no verbal communication (C003, C004 and C006), and one (C010) was too unwell on the day of the interview to participate although he was present. All of the other ten children took part in the interviews. Two of the children died during the course of the research.

The children had a diverse range of conditions. It soon became apparent that the established Together for Short Lives categorisation did not adequately reflect the individual, highly complex nature of the conditions with which the children lived. They either fell into more than one category, or did not fit into any of the categories at all.

The children participated in the interviews as much as they felt able to or wanted to. Their willingness and ability depended on factors including their clinical condition, the location of the interview, and whether or not we had met before. Only one of the children was interviewed alone, C011, who was aged 17 years and was willing to be interviewed alone at the third interview. The other children expressed a preference for the family members to be with them during the interviews. Interviews ranged in duration from 26 minutes (with a child) to 108 minutes (with a mother).

The study population is summarised in table 6.1, which details the age of each child at recruitment, whether they were male or female, used verbal or non-verbal communication, and the Together for

Short Lives category closest to their condition. The members of the family who took part in the study are also outlined together with identifying numbers for each participant. The number and location of each of the interviews are also summarised in table 6.1 below:

Table 6.1: Summary of study population.

Family	Child's age at recruitment	Male or Female	Able to take part in interview?	TfSL category	Participants and identifier	Number, location and dates of interviews
1	5 Cancer	M	Yes	1	Child (C001) Mother (M001) Father (F001)	1. Home 21.10.16 2. Ward 1.12.16 3. Home 3.5.17
2	17 Congenital	F	Yes	2/3	Child (C002) Mother (M002) Brother (B002)	1. Home 2.11.16 2. Home (M002 & C002) 14.2.17 3. Home 16.6.17 4. Home (B002) 3.11.17
3	8 Congenital	F	No (non-verbal communication)	3	Mother (M003) Father (F003)	1. Home 11.11.16 2. Home 24.1.17 3. Home 30.6.17 (interview interrupted, completed 20.7.17)
4	8 Congenital	F	No (non-verbal communication)	3	Father (F004)	1. Home 7.12.16
5	6 Congenital	M	Yes	1	Child (C006) Mother (M006)	1. Ward 2.2.17 2. Outpatients 1.6.17 3. Outpatients 12.10.17
6	18 Congenital	M	No (non-verbal communication)	4	Mother (M006)	1. Home 10.2.17 2. Home 9.5.17 3. Home 21.9.17
7	7 Cancer	M	Yes	1	Child (C007) Mother (M007) Father (F007)	1. Ward 17.2.17 2. Home 22.3.17 3. Home 11.10.17
8	5 Congenital	M	Yes	1	Child (C008) Mother (M008) Brother (B008)	1. Home (M008) 9.3.17 2. Home (C008 and B008) 8.6.17 3. Home (All 25.7.17 4. Home (M008 14.9.17)
9	11 Cancer	F	Yes	1	Child (C009) Mother (M009) Father (F009)	1. Outpatients 28.3.17 2. Ward 25.4.17 3. Home 22.9.17
10	5 Congenital	M	No (too unwell)	1/2	Mother (M010)	1. Ward 5.5.17
11	17 Congenital	F	Yes	1	Child (C011) Mother (M011) Step-father (F011)	1. Ward 17.6.17 2. Home 5.5.17 3. Home 7.9.17
12	14 Cancer	M	Yes	1	Child (C012) Mother (M012)	1. Outpatients 11.5.17

| 13 | 14 Cancer | M | Yes | 2 (as a result of 1) | Child (C013) Mother (M013) | 1. Home 15.6.17 2. Home 9.8.17 |
| 14 | 10 Congenital | M | Yes | 3 | Child (C014) Mother (M014) | 1. Home 29.6.17 2. Home 10.8.17 3. Home 4.10.17 |

Families were free to withdraw from the study at any point without giving a reason, but two families provided reasons nonetheless. One was a family where the father was the only English-speaking parent. He had a busy family life, and did not feel able to contribute further to the study due to conflicting demands on his time. For another family, their child died soon after the first interview, and his parents did not wish to continue to take part in the study following their bereavement. The third family who took part in one interview did not respond to telephone calls to arrange a follow-up interview. This was at a time when the child was about to embark on further intensive treatments, so a decision was made not to pursue the family further after three attempts to make contact. None of the families asked to withdraw the data that had already been collected from them.

Every child in the study had a life-limiting or life-threatening condition, but input from a specialist palliative care team or hospice had been an inconsistent feature of their care. Six of the children were receiving care from a specialist paediatric palliative care team or a hospice at the time of the interview. One family had experience of "palliative care" with their child, who had been expected to die in the neonatal period. They had been discharged home soon after birth "for palliative care" but no specialist services were ever involved. Their child was expected to live for only days or weeks, but had survived and was six years old at the time of interview. Another child had never received a referral to specialist palliative care services as they had never been considered unwell enough, according to their family, despite having had a number of life-threatening deteriorations. The involvement of palliative care services with each member of the study population is summarised in Table 6.2 below:

Table 6.2: The involvement of palliative care services with the study population

Family	Child's age at recruitment	TfSL category	Involvement of a palliative care service?	Details
1	5 Cancer	1	No	Never offered. Plan was for curative oncology treatment.
2	17 Congenital	2/3	Yes	Referred to local children's hospice early in childhood. Also referred to community palliative care team. Known to both services for years. Referred at a time that was thought to be

				the end of the child's life, in intensive care. Advance Care Plan in place, updated annually.
3	8 Congenital	3	Yes	M003 referred herself to the service having been bereaved of a child with the same condition as C003 in the past. Well known to community palliative care team. Advance Care Plan in place, updated annually.
4	8 Congenital	3	Yes	Children's hospice provided respite care. Community children's nurse involved. Referred by GP. No Advance Care Plan.
5	6 Congenital	1	No	M005 and C005 talked openly about the life-limiting nature of the C005's condition and uncertainty about the future. Frequent contact with hospital teams but not referred to a palliative care service.
6	18 Congenital	4	No	M006 aware of palliative care services but felt that C006 had never been considered unwell enough by his clinical teams for a palliative care referral. Received respite care from another provider (via social services). Also received personal health budget.
7	7 Cancer	1	No	Plan for curative oncology treatment. Although the family were aware that this may not result in cure, they had not received a referral to palliative care services and seemed to consider this as a distinct element of care that was instead of active treatment, not alongside.
8	5 Congenital	1	No	Had been discharged as a neonate for "palliative care" but did not die. No specialist services involved at the time. No Advance Care Plan.
9	11 Cancer	1	No	Consultant had tried to engage family in conversation during an ICU admission – unclear whether palliative care referral was offered. Complicated situation, with delayed diagnosis.
10	5 Congenital	1/2	Yes	Referred to hospital palliative care team for pain management. No Advance Care Plan.
11	17 Congenital	1	No	Family aware of palliative care services as worked in social care. Had not been offered referral. No Advance Care Plan.
12	14 Cancer	1	No	Not known. Plan was for curative oncology treatment.
13	14 Cancer	2 (as a result of 1)	Yes	Referred to community palliative care nursing team for monthly injections. Known to hospital palliative care nurse. No Advance Care Plan. M013 found the word "palliative" very difficult to talk about.
14	10 Congenital	3	Yes	On waiting list for respite at local children's hospice. Referred to community palliative care team on discharge from hospital. No Advance Care Plan but discussions had in hospital and the community, and were considering.

Recruitment: Focus groups

Focus group discussions were conducted at paediatric palliative care network meetings in four sites across England (West Midlands, London, Yorkshire and Humber and the South West). The first focus group was carried out on the 4th December 2017, with the others being carried out over the next six months on 15th March 2018, 9th May 2018 and 29th June 2018.

Study population: Focus groups

The focus groups were conducted with healthcare professionals from a range of backgrounds (medical, nursing and allied healthcare professionals) who all had an interest, involvement or experience of providing palliative care to children. A total of 86 healthcare professionals consented to participate; 71 took part in the focus group discussions. 16 were doctors (consultants or registrars) from paediatric palliative medicine, oncology, intensive care, general paediatrics or community paediatrics. 50 were nurses from a range of specialities including paediatric palliative care, paediatric oncology and children's community nursing. Those who were not from a medical or nursing background included play therapists, hospice managers, pharmacists and clinical psychologists (hereafter referred to as Allied Healthcare Professionals, AHPs). Table 6.3 summarises the focus group participants.

Table 6.3: Focus group participants

Focus Group	Date	Participants		
		Doctors	Nurses	Other
1	4.12.17	4	20	0
2	15.3.18	4	16	1
3	9.5.18	4	4	1
4	29.6.18	4	10	2

The approach to recruitment allowed for a large number of healthcare professionals to participate, at a time that was convenient to them. The approach resulted in the conduct of focus group discussions with relatively large numbers of people, however the discussion seemed open and positive feedback was received informally via email following each focus group.

Opportunity was provided for network members not to participate if they did not wish to. It is possible that given the format of holding the focus group during network meetings, some will have felt it was difficult to leave the room if they did not wish to participate. This was a potential limitation of the recruitment that should be considered in more detail for future studies.

6.3. Summary of Chapter

This chapter has outlined the recruitment strategy and challenges encountered. The recruitment targets were met, but there were strengths and limitations to the strategy that have an impact on the findings and interpretation of the research. Recruitment of children to palliative care studies is an under-reported area of practice, there is a need to share learning from recruitment more widely.

7. Findings 1: The child and family situation, micro-system level findings

7.1. Overview of Chapter 7

Chapter 7 provides the results of the thematic analysis relating to the child and family experience of life with a life-limiting or life-threatening condition and interactions with healthcare at an interpersonal (micro-system) level. The micro-system refers to the child and family's immediate relationships with individual healthcare professionals. As outlined in the methods (chapter 5, section 5.4), thematic analysis was carried out firstly on the child and family interview data, followed by the focus group transcripts, with focus group data used to provide further insights into the experiences and perceptions of children and their families.

The findings provide evidence to address the research questions:

- What are the lived experiences of children with life-limiting and life-threatening conditions and their families?

- How do children with life-limiting and life-threatening conditions and their family members perceive healthcare services, and in particular "palliative care"?

Quotes to illustrate the findings are presented, with "SM" referring to me as the researcher. The abbreviation "Res" refers to focus group respondents.

7.2. Life with a life-limiting or life-threatening condition

The analysis revealed four interconnected themes, which closely interact with each other in the lives of the children and their families:

1. A life-limiting or life-threatening condition in a child permanently alters the family story.
2. A fluctuating trajectory shapes the family's hopes and expectations.
3. The child does not wish to be defined by their condition.
4. Death is a constant presence.

Theme 1: A life-limiting or life-threatening condition in a child permanently alters the family story

The onset of a life-limiting or life-threatening condition in a child significantly and permanently changed the family story. Families had to quickly adapt to their new situation, rapidly altering their expectations of both the immediate future and their longer-term hopes and plans. The realisation that the child's condition was life-limiting or life-threatening brought with it an often implicit, unspoken, knowledge of the possibility that the child could die. This was a situation that was against the natural order of life and against societal norms. It was an intensely emotional time:

Mother: But there is an expectation that the parents will pass away and not your children, yeah …
You should never have to bury your child…
(Interview 2, M006)

For the five children with cancer, the point of diagnosis was a shock ("*it hit me like a tidal wave*"

M001) and admission to hospital for treatment occurred within hours or days:

Mother: And she [the doctor] said you need to prepare for the worst. It's either this one, or this one.
And we were like, it still didn't make any difference to me really, you've still got that word
[cancer] in your head … The next 48 hours were kind of manic. People coming at us from all
angles … And then 48 hours later we were… you'd had your line fitted hadn't you [C012]?
(Interview 1, C012 M012)

This was different to the nine children who had congenital conditions, where realisation about the

nature of the condition had been more gradual:

Mother: No, no he literally he was born, and everything kind of like went from whoa, hang on what's
happening here … So he come out, cried, and then stopped, and that was it, erm he had to
have a whole lot of like nitric oxide, cPAP, he was on ventilation for, ooh, a few weeks at least
… yeah, so it was just like, it all kind of went downhill, like oooh what's going on? You know,
because they didn't pick up anything during pregnancy.
(Interview 1, C005 M005)

In both situations, a period of initial shock and adjustment was followed by realisation relating to the

severity, extent and implications of the condition. The children and their families embarked on a

lifelong process of learning, focussed particularly on recognising and managing changes in the child's

condition and the plans for treatment. A concurrent, continual process of adaptation occurred, with

children and family members having to frame then re-frame their hopes and expectations of what

might happen on a day-to-day and longer-term basis. For many of the families, the child's condition

became a family vocation, with family members taking on caring and nursing roles. The family home

also became a healthcare environment. The demands on family members were significant, and

although they expressed a determination to "get on with it" this was not a situation of choice. These

findings are presented here as three sub-themes:

Subtheme 1: The child's condition becomes a family vocation
Over time, the child's condition and management of their care became a way of life for families, a

"full time job":

M014: Ultimately, to me, he's my responsibility and he trusts me so implicitly because I've always
managed everything. But now the scale is just so vast that it is a full time job.
(Interview 1, M014)

From the children's perspectives, their parents were regarded as their trusted representatives, decision makers and spokespeople; *"the best parents in the world"* (C009). The children and their families embarked on a process of learning related to the child's condition. This initially took place through the provision of verbal or written information from healthcare professionals, which was not always easy for family members to understand and retain. Often a large volume of information was provided, to a parent who was also trying to come to terms with their new circumstances, support their child and provide information to other family members:

Mother: And they were asking like, when we take the blood can we have your leftover samples and stuff. And I'm like hang on a sec, I'm still trying to get used to [the diagnosis]… and then obviously family are like messaging, and I'm trying to sit there, and my husband's buried his head in the sand because he doesn't like hospitals and stuff anyway. So I'm trying to take on board all this information, and I'm like …
SM: Yeah. Do you get any of it written down?
Mother: Oh yeah, booklets, and booklets, and booklets. And they're like read that when you've got five minutes, you don't get five minutes. You just kind of, you have to jump in and learn it as fast as you can.
(Interview 1, M012 C012)

Family members described how valuable it was to have professionals who tried to help, by providing information more gradually over time:

Mother: …you know Dr [name] came to show us this [treatment programme] probably in that first week, maybe in the second week, cos he tried to sort of drip feed, cos there is a lot, to take in, and when you're shocked by the diagnosis in the first place…
(Interview 1, M001)

The children's conditions were characterised by complexity related either to the condition itself, the treatments, or the associated symptoms. Over time, family members became experts in managing not only this complexity, but their individual child's needs within that level of complexity. They developed an in-depth knowledge of the implications of medication changes and test results. In the example below, a father was listing the number of important considerations that he expected to be addressed before his daughter could be discharged from hospital:

Father: We'll be out maybe, they've said definitely over the weekend, hopefully not too deep into next week but depends if they get the warfarin sorted, and the potassium. And the creatinine, and the oxygen at night, and the blood pressure and the magnesium…
(Field notes, Interview 1 C011, F011)

For some children, their medical complexity included living with medical technology, such as enteral feeding (via a percutaneous endoscopic gastrostomy (PEG), or total parenteral nutrition (TPN) via a central line), the delivery of intravenous fluids and medication via PEG or central line, or

trachoestomies and long term ventilation (LTV). Family members managed these medical

interventions at home. One mother and child described their daily routine as follows:

Mother: So 6 o'clock, well [C002], you are mostly snoring when I'm scrubbed up. So you have to be
sterile, so you have to be scrubbed up. You have to scrub up and be sterile, so
Child: About 50 times
Mother: She says, it's about 50 times a day, so then I do that and then at 8.00 I start re-scrubbing up
and getting everything ready for 9,
Child: Again
Mother: And then at 9.00 you do some more meds, and then I'll flush her off her PN. Then I put her
on IV fluids. So you calculate what she's left, had left. So she's on that until she's had
whatever she needs to have. Some days she can be on it for eight hours, some days she can
be on it for three hours. So then you re-scrub again and flush her off at the 12.00s, they can
keep on going into the line, so you can give the 12.00 meds. Then at 4.00 I do her PN, scrub
up, do her PN and do her other lots of meds
Child: oooh. And Daddy, he can set them up
Mother: Then again at 6, then again at 9. And the last one is 1.00 in the morning. So we have 1 til 6
sleep, but then you're checking her temperature cos of her having sepsis and TPN, she can
get a line infection. So you're taking her temperature 7, 8 times a day.
You just get used to the routine. She tells me to get lost some days cos I'm at her like all the
time.
(Interview 1, M002 C002)

Subtheme 2: Home becomes a healthcare system
The children "loved" to be at home: "*I love being at home … being in my own bed*" (C009). Home

became a healthcare system of its own with necessary adaptations to the house including ramps to

the front door and handrails. For some, the adaptations were extensive, including ground floor

extensions with new facilities such as a bathroom with hoists, or lifts to upstairs floors. Equipment

was present including hospital beds, oxygen concentrators, drip stands and in one case a number of

ventilators. Three of the families had built sheds in the garden for the storage of medical supplies:

Mother: Oh there's more than that. The shed's terrible.
SM: So your shed is full of your monthly supplies?
Mother: Yeah. Nappies, stoma stuff, catheter stuff, everything … we had to buy the shed because
the house isn't big enough for all her stuff.
 (Interview 2, M002)

The impact of the child's condition on the home environment was striking, even for experienced

healthcare professionals. The adaptations that had been made at the home of C014 in order to

accommodate his complex needs had shocked and upset his community nurse:

Mother: My community nurse, she came, saw him at home, saw everything there the first time she
came, and when she came the second time, she said, "I've got a confession", she said, "I
cried when I left here. She said "it's like ICU in there". And it is. It's PICU at home. And it's
the pressure of PICU at home.
(Interview 1, M014)

Parents were responsible for the design and management of the home system. They constantly negotiated and re-negotiated with the other systems around them, including the health and social care systems. There were other important but perhaps less considered systems to be negotiated, such as the regular bin collection and grocery shopping:

Mother: Even something as simple as… it affects everything. I've had two new bins because the rubbish that we're producing and all the recycling I'm doing, just down to that, the one day I had no milk and the bin man wouldn't take my bag of rubbish because it wasn't in a proper bin. And it's just something like that… I just came and I sat at that dining table and I cried.
(Interview 1, M014)

Parents had their own systems such as charts and calendars in place to monitor stocks of medical supplies and feeds. One family had converted an entire bedroom to storing notes and documents related to their child's condition and care:

Mother: I, my small room plus junk room is absolutely full of folders, contacts, leaflets of every organisation available, you know, keeping up to date with things, and yeah, I have a lot
SM: Can you show me?
Mother: Yeah, you'll have to excuse the mess
SM & M003 walk upstairs to small room which is full of bookcases containing folders and boxes of notes.
Mother: This, so I've got all this, I've got a filing cabinet, and I've got literally got paperwork here, I've got paperwork in these boxes, I've got a lot in the attic, I've got folders there, got paperwork here, I've got stuff in there..
(Interview 1, M003)

Family members spoke about how difficult it was to manage the day-to-day practicalities and logistics of caring for their child. They carried on with determination, but this was not a situation that they had chosen, and this brought other challenges.

Subtheme 3: The situation is not one of choice:
Healthcare professionals in the focus groups recognised the demands placed upon the family:

Res: Nobody in their right minds would choose to have a child who is dying
(Nurse respondent, focus group 2)

Parents described the significant change in their parental role that occurred when their child became unwell, with at least one parent taking on the role of carer. Some perceived this change as a threat to their personal identity, and described how difficult it could be to accept that this was their role:

Mother: … I don't mind, but sometimes, you know. But that's my job isn't it? I think that's my job. I get carers [allowance], that's my job. I know it's not a job looking after your own kid, but… I feel horrible, that's horrible, that's hard to deal with.
(Interview 1, M013)

Family members expressed their feelings of disempowerment and intense vulnerability related to their child being seriously unwell. They depended on healthcare professionals to make key decisions that affected their family life. In the example below, one mother (M007) described learning to manage the lack of control that she had over decisions about going home while her child (C007) had cancer treatment:

Mother: *It's so painful at the time you just go through the motions really, don't you … it's horrible the fact that you have no control over what's going on with your child. It's like on Saturday, we felt as parents that he was well enough to go home and, we'd never take him home if we didn't think so. But the thought of taking him home after the [treatment] was frightening enough. But because you're not given any of that control, it's taken away from you, so learning to do all that is really hard.*
(Interview 3, M007)

They described a position of passivity in terms of decisions about their children's medical treatment; "*all along, we just have to go along with all they say*" (F009). With no control of either the disease or the treatment plan, parents described feeling like bystanders. They had no choice but to trust their healthcare professionals.

Mother: *You just trust these people, not with your life, with his life, almost, and there is nothing that I can do other than being there, and holding his hand and offering reassurance and loving him. And I do very easily, and thankfully hand over all of the other stuff, to the people that know best. Erm, yeah, but you know, it's not optional, whether to trust them or not, I have to.*
(Interview 1, M001)

Mother: *It is, the way I feel is that, at the minute, we're watching him be tortured, and we're just standing there, doing nothing. That's how I feel, because he's in pain all the time.*
(Interview 1, M007)

This passivity was particularly difficult when the children developed distressing symptoms. Pain was the symptom most frequently talked about during the interviews. Witnessing their children in pain was a major cause of distress, concern and anxiety for parents, who described a feeling of helplessness. In the example below, pain relief medication had run out during a scan:

Mother: *He [C001] was in a lot of pain. I was just like I'm really sorry this is really upsetting me, how much longer are we going to be? He [radiographer] was like "umm, about 8 minutes". I was like "ok, 8 minutes is ok, but let's just be as quick as we can", and then, so we got to the end of it, and then the PCA had run out. So there was this funny, it said disabled or something, and I looked at the nurse and I said "ooh, what's happened, something". And they sort of looked at each other and said, "it's run out". And at the time I was like "Oh right ok", and then of course by the time we got, so the porter had to come down and get us from the CT scan, then we had to get back up to the room, and by the time they'd sorted it all out, another 45 minutes, an hour, so probably about 2 hours later, without pain relief. So this was the second night of pain …*

(Interview 1, M001)

Family members recognised that there was often a psychological element to their child's pain, but felt that these psychological concerns were not always addressed: "*they do a lot, obviously, medically, of fixing the children, but in their heads, not so much.*" (M007). They perceived that healthcare professionals placed emphasis on physical causes of pain, with attempts made to provide medical explanations and subsequent drug treatment, rather than addressing psychological concerns:

Mother: But then following the chemo he's had what they think is mucositis. So he's in pain now in his chest, his stomach. It was very bad in his mouth. But they haven't really got to the bottom of the pain as such.
Father: He's still got pain.
SM: Okay.
Mother: Some of it is psychological, but he has definitely got pain.
(Interview 1, M007 F007)

The roles of parent and carer could be exhausting for parents but they generally expressed a determination to "*get on with it*". This was difficult to maintain at times, as described below:

Mother: And I had to look in the mirror and just keep saying, stop it, you know, you've got to eat, you've got to stop, because if you aren't there for him no one else, it's not you that's bad. I don't know how I've done it but I had to box it all and just do it, and like it was really hard.
(Interview 1, M013)

Mother: I think everyone's got different abilities. Thankfully, I am an organiser. We only had this conversation… this conversation was raised twice yesterday, one by my CCN in front of her manager … And then, my mum came for tea and she just said to my [C014's] carer, she was sat the table, so I fed the carer as well. And you know, she just said, "what would happen if you weren't you?"
(Interview 2, M014)

C014, the child of the mother in the second quote expressed an awareness of the impact that caring for him had on her, and had a desire to protect her. He had offered to go back into hospital a few days after discharge because he recognised how tired his mother was:

Mother: [C014] is worried about me being too tired to look after him without making a mistake. I'm sad that my 10 year old is worrying about me! He actually said he wanted to go back to [hospital] so I could get a rest and then he would come home again.
(M014, field notes from interview 1)

Theme 2: A fluctuating trajectory shapes the family's hopes and expectations

All of the children and their families lived with clinical uncertainty, not knowing what would happen next in terms of the course of their condition, effects of treatments or new symptoms. Their

conditions, whether they had an oncology diagnosis or non-malignant disease, were characterised by periods of stability, usually for weeks or months, punctuated by sudden, acute and often unexpected deteriorations. A period of recovery followed, with adjustment to a new situation, sometimes adapting to a new treatment regime or medical technology. Experiences such as inpatient treatments, surgery, or an admission to the intensive care unit, which at first were alien and frightening to the families, became normal as they happened more frequently. Long hospital stays through a series of fluctuations in the child's condition were a common occurrence; three of the families described hospital stays of over 10 months at a time. An older brother described the fluctuations in his sister's condition as living like a "*human yoyo*" (B002). Others used the phrases "*ups and downs*" or "*ins and outs* [of hospital]".

Mother:… Every two or three weeks and we're back in again. He's constantly getting [infections]
Child: In and out, in and out, in and out
Mother: In and out, yeah
SM: Do you feel very poorly when you have an infection?
Child: I feel like crying when I come in
(Interview 1, M005 C005)

Symptoms associated with the child's condition or treatments, such as pain or nausea and vomiting, also tended to fluctuate and change regularly. This added to the complexity of the child's condition. With every deterioration or change in the child's condition or treatment, there was a further process of learning and adaptation to contend with. Coming to terms with each change as it became apparent was difficult, as described by a mother in the quote below:

Mother: Yeah, you know one thing, one thing, it's easy to cope. When he was one, we thought, we thought even with [organ problem], we thought, ok the maximum he can have is him having a [organ] transplant and thats it. You don't think even transplant can be really complex. And er, we were like mentally ready for him to be on [long term treatment], have the transplant, and er, then we found out that his [another organ] is no good, you know? And, erm and it needs to be a combined transplant. Took us ages to have that in our mind that he needs both. Then we like, when we were like mentally ready for him to have both, [two organs], then they told us that he's got a problem with his [another organ].
 (Interview 1, M005)

The children who had experienced life-threatening deteriorations in their condition requiring emergency admission to the paediatric intensive care unit (PICU). They described these admissions as "*freaky*" (C007), "*scary*" and "*frightening*" (C002). One child described realising how unwell the other children on PICU were, undergoing surgery in their PICU beds, and being unable to eat:

Child: They don't, like, move them to the theatres they just do it in the beds.
SM: Yeah? Did you have to see any of that?

Child: *No. In PICU you don't order food.*
SM: *You don't order your food, right. How come you don't order your food?*
Child: *Because most kids wouldn't eat in PICU.*
(Interview 3, C007)

Factors that were important to the children during their PICU admission did not tend to relate to their condition, but more to feeling that their personal needs and priorities were met. One child described the importance of having a games console for entertainment and a mobile phone that he could use to contact his parents. Another child described the importance of being able to communicate her need for pain relief on PICU while she was ventilated:

Child: *Yeah, I couldn't talk*
Mother: So what, because you couldn't talk, what were you doing?
Child: *Signing*
Mother: Signing to everyone, wasn't you
SM: *Did they understand?*
Mother: Yes. And they was really helpful when you were signing to them
Child: *I was like please can I have my [pain relief] spray, and they gave it to me*
(Interview 2, C002, M002)

Over time, intensive treatments, admission to PICU and recovery from significant deteriorations was expected; families acclimatised to this course of events and accepted it as part of life with their child's condition. The children seemed pragmatic about the severity of their condition at these times. One child stated "*I don't mind… It just happens with me";* she considered these episodes to be part of her day-to-day life, only worrying if she "*thought about it a lot*" (C011). Families described admissions to PICU becoming less scary as they happened more frequently, even becoming "*a bit cocky*" (M007) about PICU:

SM: *Yeah. Because you'd been to ICU before, hadn't you?*
Father: *Yeah. Way back at the beginning when he was first diagnosed, yeah.*
SM: *Yeah.*
Father: *It wasn't as scary this time round, was it? Still not a very nice place to be in, with all those different people coming in all the time.*
(Interview 3, F009 M009 C009)

Mother: *But then we were a bit cocky about ICU. We went "yeah ICU, it's fine".*
SM: *Because you had been before had you?*
Mother: *… Oh been there, done that now. And then he went downhill again and they took him down to ICU and it was just awful. He couldn't breathe.*
(Interview 3, M007 C007)

Several of the children had made one or more "miracle recoveries" from life-threatening episodes. Families were described by professionals as *"being able to grieve over and over again"* (nurse, focus

group 4) in relation to these experiences. Family members expressed hopes for the future, based on the premise that this fluctuating condition trajectory would continue or improve:

M002:   *I've lost count of how many times they have said it's the end.*
(Field notes, M002)

Mother: [I] sent her [the doctor] the end of treatment picture, and she just sent back, "I'm just so… that has just made my day". And she said, "he's the example I use when other families come in and they go through tough times", she said, "because he's the miracle one". … Mum overheard [the doctor] saying to others, its the biggest turnaround he's ever seen.
Child: Ever. Biggest turnaround ever.
Mother: And he's had no problems following as such, he's still on oxygen and stuff overnight but I don't think that will be long.
(Interview 3, M007 C007)

Some spoke more openly about the long-term implications of the child's condition, recognising that while recovery from acute deteriorations was expected, there was no cure for the child's condition. They voiced concerns about the implications this would have on their child's life, and how they would discuss this with them in the future:

M008:   *We've got to tell [C008] at some point she's got a life-limiting illness, she doesn't know. She knows she's had a [organ] transplant, and she knows that she's got a button and she's different, but she doesn't know that she's not going to be a granny, or things like that.*
(Interview 1, M008)

Theme 3: The child does not wish to be defined by their condition

The children who took part in the study tended to be pragmatic about their conditions, accepting them as part of their life. They demonstrated a detailed knowledge of their conditions, regardless of their age. They did not wish to be defined by their condition and had many more ideas and interests to discuss during the interviews than the implications of their condition or experiences of healthcare. The younger children displayed their understanding of their condition through play:

Mother: All of [C008]'s play, if she's playing babies, revolves around hospital things. She's got a button [PEG tube] for her baby. They had to make an incision into the baby's stomach, there's a hole in there where she can put a button, and she's got a pump and a feeding tube and she puts the backpack on and takes the baby for a walk, all of the things that happen to her.
(Interview 2, M008)

The children were aware of both acute and insidious changes in their condition and occasions where those changes required attention, intervention by a carer or further medical assessment:

Child: Yeah, my hand kept on like going like weird, and then I tried to write and it just kept on going in this funny position every time, the same position and she just… my mum just said, "oh it's nothing".

Mother: You wouldn't know would you with all of his other problems.
SM: Did you think she was wrong, or right?
Child: No, I… because people say like calcium all the time, I got cramp and then I was just… I didn't really think about it.
Mother: And then I… we went to the… where did we go now, it got quite bad didn't it?
Child: It was like in my legs.
Mother: Yeah, and then he was like… because obviously then we took him [to hospital] and they were like, oh my god [there was a diagnosis].
(Interview 2, C013 M013)

Child: Mummy…
Mother: Yeah?
Child: Bagging…
Mother: Bagging? Okay, sorry, let me just give [carer] a shout. I think he needs to do some bagging [C014]. Right, [C014], tip yourself back then please sweetheart.
[Mother attends to child's care, SM leaves the room at child's request]
(Interview 2, M014)

The children were also aware of changes in their medication. In the example below, C007, who was seven years old at the time of the interview, had been more aware of a new medication than his parents:

Father: Its like this morning, with the tablets. I said what are those two tablets? Because I'd never seen these two tablets. And he said they go under my tongue. And I'd never seen them. … And he only started them yesterday.
Mother: Yeah, but she [nurse] said he'd had them before, and I said he hasn't had them before.
Father: He's never had those before, never. But he knew. … he knew what they were for, and I didn't.
(Interview 1, M007 F007 C007)

Some of the children were aware of the implications of clinical measurements such as oxygen saturations, particularly if this was monitored at home, for example alongside long-term ventilation. They came to rely on these measurements for reassurance and a feeling of safety:

Mother: He knows what it means to be safe. He knows things have to be changed regularly, he knows what his numbers should be, he'll say, well that oxygen cylinder's run out because I can't taste it anymore. And we'll look and it has.
(Interview 2, M014)

Despite being passive recipients of treatment plans and unpredictable fluctuations in their condition, over which they had no control, the children sometimes felt responsible and wished to protect their parents (C009, for example, had apologised to his parents for a collapsing at home).

The children did not wish to be defined by their conditions, despite the impact on their health, lifestyle, family, home and personal appearance. Their descriptions of living with their conditions were dominated by their own priorities for life, and the impact that their conditions had on the

things that they wanted to do or achieve; *"Stay alive, and be happy. That's the aim"* explained one of the children (C014). They talked more about their own interests and the activities that they enjoyed, such as seeing friends, going to school and going on holiday; *"just want*[ing] *to be normal"* (M013). They expressed a desire to take part in activities with other children of their age such as going to school or going for *"a walk in the village and going to the "All you can eat" buffet"* (C014). Their medical conditions significantly affected whether or not they could take part in these activities:

Child: The things I like doing I sometimes can't, but the things that I don't like doing I can.
SM: So tell me a bit more about that?
Child: Like there's swimming, performing arts, I think, like everything…
Father: Yeah. Yes, up until recently for her condition and the issues she has, she's always been quite active. She goes to a special school in [town] for people with learning disabilities, and you always used to love going on the trips didn't you, doing the canoeing, the abseiling.
Child: Yeah, like the canoeing, the abseiling, I can't do because of my [organ transplant].
(Interview 1, C011 F011)

SM: You mentioned he went for a day at school
Mother: Oh yeah, he was not, only a day, it was only three hours. He was fine, he was so happy. He went on Wednesday. He was telling everyone that "oh I went to school" and he was looking forward yesterday to go but … [sudden decline in health]
(Interview 1, M010)

13 of the children lived with medical technology or equipment that altered their appearance, including Hickman lines, nasogastric tubes (NGT) and PEG tubes. Some had scars and stomas. 12 used wheelchairs to aid with mobility, four of these children were wheelchair-dependent. One of the children had a tracheostomy and lived with long-term ventilation. These external markers of illness sometimes attracted the unwanted attention of others. The children engaged in decisions that involved weighing up the risks and benefits of each of these interventions, and how to manage them on a day-to-day basis, where possible:

Child: [I'm] forced to have a tube [NGT] in … in my nose. So she [another young person] was being mean. She was like "ugh, up your nose",
Mother: And what does the tube help with?
Child: The pain
Mother: … So what did we say? If it makes you that upset we can take it out, and what did you say?
Child: No
Mother: No, because why did you say no?
Child: Make more agony
Mother: It would make you in agony, clever girl. So you've, So you've made the decision that you want to feel better and it doesn't matter what anybody else thinks does it.
(Interview 2, C002, M002)

Mother: Yeah. When she first started school, it was a bit of a novelty, people wanting to see her button [gastrostomy] … times when we're at swimming, if she sees someone staring at her,

she'll be like giving them a right glare. I'm like "don't do that, they're just, because it's different they're just having a look".
(Interview 2, M008)

The subjects that the children were not willing to talk about during the interviews were as notable as the subjects that they did wish to discuss. A common finding throughout the interviews was that they would divert the discussion away from healthcare-related topics to other completely unrelated subjects, or an activity, including those that had been designed to aid the interview process. In the example below, C008 diverted the conversation to the felt tip pens that were intended for use in a draw-write-tell exercise:

SM: You have to go [to hospital] quite a lot, yeah. So is it, erm, how do you feel when you see your doctor?
Child: Happy
SM: You feel happy. What does she do?
Child: (referring to felt tip pens) They are the colours I like
SM: They are the colours you like, right ho.
Child: These aren't in the rainbow
(Interview 1, C008, M008)

On occasions during the interviews, the children would ignore questions about their health and healthcare, or decline completely to talk about their medical condition. This often occurred at points where the interview touched on significant moments in their lives, such as a serious deterioration in their health or a hospital admission:

C014:   "there are times in my life I don't want to talk about, like [the experience of a cardiac arrest]" (Interview 2, C014).

The children described moments when they felt that their views and concerns were unheard or unaddressed. This applied both to their health and to their other priorities. A notable example was one of the children at the time of diagnosis. Following several GP appointments, one child, C007, insisted to his parents that something was wrong and that he needed to be seen in hospital. He was diagnosed with cancer following this accident and emergency (A&E) attendance:

M007:   We kept thinking "why won't the doctor just give him some antibiotics", because obviously he's not getting any better. That carried on and on and on. And then finally he said to us "we need to go to the hospital now". And we took him to A&E then.
(Interview 1, M007, C007)

The children's priorities also went unheard, for example, wanting to play outside. In the example below, a child who was seriously ill and bedbound for the duration of the study, expressed his wish to go out and play. The request came in the middle of a conversation about his health, and was not

acknowledged in any way by the adults in the room (including me), so he was not taken outside to play:

Mother: Yeah, they need to get back on top of it, you know, I think [physiotherapist]'s going to sort him out today. They're liaising with his school because he's got a physio at school so they're going to come out to the house when we're at home. The physios will sort him out when he's in here. I said while he's in here and doing nothing they could get him down the gym and that, doing stuff.
Child: Can I play out in a bit? I love that.
Mother: They come up some times and he's attached to fluid so he's restricted to go anywhere. But while he's not he can go down.
(Interview 2, M009 C009)

The children expressed anger and frustration when they felt that their, often expert, knowledge of their condition was not acknowledged by healthcare professionals, or when their personal priorities were ignored. They disengaged with healthcare professionals and found it difficult to trust them. This was a particular problem if they had not met these professionals before. In the example below, C007 was describing a recent experience in the Emergency Department (ED). The experience contrasted with C007's experience of care from his specialist medical team:

Child: They don't even know what to do.
Mother: What don't they know
Child: How to care.
SM: How to care?
Child: Once they didn't even know how to put a cannula in.
SM: Really? Do they know all about your treatment and things do you think?
Child: No.
SM: No. So do you have to tell them?
Child: No. I said to have a guess.
(Interview 2, M007 C007)

The healthcare professionals who took part in the focus groups also recognised this situation. They were able to provide numerous examples of moments where the child's needs and priorities seemed to be unaddressed by their healthcare team. The example below describes a child perceiving that she was being cared for "as a disease" rather than as a person during a hospital stay:

Res 1: "You can talk to me, you know you don't need to look at me as a disease, you can look at me as a person". Erm, all of that came out. And actually the staff found it really difficult on the ward because that young person was obviously really cross about some of the things that were being said to them, and then they sort of picked out the best nurses, and would only have those nurses because they were the ones that would relate to them, erm, and I [children's hospice worker] had to go in and I had to tell the staff that actually this was quite normal for this young person, and actually you know they should be listened to, what they had to say and actually [child] wasn't being difficult, she was just cross, because she wasn't being listened to. And she wasn't having her needs met.

The feeling reported by children and their families of not being heard is considered further in the second findings chapter. Insights into the child and family interaction with the healthcare system go some way to explaining why they often feel unheard. A further relevant and important factor was the life-limiting or life-threatening nature of their condition, and the often unspoken possibility that the child may die, which is explored further in the following sections.

Theme 4: Death is a constant presence

The children's diagnoses and each significant deterioration in their condition were both associated with an awareness amongst family members that the child's life was threatened and fragile. Nine of the families spoke openly about their knowledge of the life-threatening nature of their child's condition during the interviews. The accounts of families of children with an oncology diagnosis differed to those with congenital conditions.

The children with cancer and their family members described a series of distinct events, starting with the child becoming unwell, and the diagnosis of cancer being made. The diagnosis of cancer was immediately considered life-threatening, but the possibility that the child may die was acknowledged to varying degrees by family members. Medical management plans focussed on curative treatment options, which started very soon after diagnosis. There was not much opportunity to dwell upon the possibility that the child may die. Sometimes it was only after a significant deterioration in the child's health that the life-threatening nature of the episode was realised. One mother, whose child had cancer, spoke about her retrospective realisation of how seriously unwell her child had been when he was admitted to PICU. She described a change in her coping strategies, with denial about how serious the episode of critical illness was at the time, and not thinking about the future anymore:

Mother: *And like I say I can always protect my… a coping mechanism for me is almost a kind of a, it didn't really happen or it wasn't that bad … And everything points in fact that actually it [the admission to PICU] was pretty big and pretty bad, but I don't want it to be. … But what am I scared of? You know, acknowledging that it was bad.*
SM: *Do you think about the future as well?*
Mother: *If I think it's too bad then I get upset and I'm trying to not get upset. But then, I don't worry about the future anymore.*
(Interview 2, M001)

The family members of the children with congenital conditions, including those who had been recipients of organ transplants, acknowledged the life-limiting nature of their child's condition more openly than those with an oncology diagnosis. One mother, who had already been bereaved of two

children, described having the thought that there was a possibility of her child dying as "*always in the back of my mind*". She described how she coped with this:

Mother: I've never been able to think, they're not going to get better because I think I'd get really quite down. I mean we've had… they've had friends who have passed away from chest infections and things like that, who have gone to hospital and not come home. And when that happens you always think, well that could be me. And you don't want to think it could be me otherwise it would cloud your whole day, your year, everything. So you always have to be that positive, no they're going to get better. And it is always in… it's in the back of your mind, but you really have to shove it right to the back of your mind, like I have when I've been to too many funerals of their [the children's] friends to think too much about it. You have to be positive, it's the only way you cannot be depressed is to think it will never happen.

(Interview 2, M006)

Another mother described a particular time when she had been truly concerned that her child would die. This conversation took place after the interview, when the voice recorder was turned off. She explained that this was the first time she had spoken about it and that she had found it too difficult to discuss with anyone previously, including close family members. A moment when her daughter was taken for emergency surgery was the "*first time I've ever really worried that it was end of life*", despite a number of previous very serious deteriorations in her child's health, admissions to PICU and a hospice referral for end of life care. She described sitting alone, waiting for her child to return to PICU from the operating theatre and feeling "*petrified*" (Mother 002, Interview 2, field notes).

Descriptions of open discussions with healthcare professionals about the possibility that a child may die were rare. This was despite the significant life-threatening deteriorations that their child had experienced, and, for some, despite experiencing the death of other children who had become friends on the ward, as described by M012 below:

Mother: But then like there's families coming in here, they'd lost children, they were bringing gifts to the children, and that would tip me over. And then I'd start crying again …   that would set me off, and our families, and friends, and staff were donating games and stuff for their kids on the ward, that would start me off … You have, we have, [C012] does have kind of like wobblies. Some of the medications have caused, it's not, you've only cried like really twice haven't you. Diagnosis and a few little bits since. Our friends lost their little one two months ago while we were in here, and we had a bit of a cry over that, didn't we.

(Interview 1, M012, C012)

Two subthemes were identified within the theme that death was a constant presence. Firstly, that the children had an awareness of death, and secondly that there was a feeling amongst family members that time with their child may be limited.

The children had often disclosed their awareness of the life-threatening nature of their conditions to their family members. Some had voiced concerns about the fact that they might die. One of the children had confided in his mother after an admission to PICU, that at the time of the admission he had thought he was going to die:

Mother: He said "I thought I was going to die". And I said "alright, okay". I said "when was that then?" And stupidly I almost put words into his mouth, I said "oh in hospital". As soon as I said it I thought shut up and just let him talk. I said "oh, in hospital". And he said "yeah, when my tummy was so sore". I said "yeah" … yeah, so he said, he said that, "when my tummy was so sore". And then he said, "you know", he said "I'm really glad I didn't". You know, sort of really flippant, and really sort of… and I just looked at him and I was like, I don't know my eyes were like, I didn't cry as such, but my eyes were welling up, and I just gave him a massive hug. And I said "we're really glad you didn't too". Thinking bloody hell, massive understatement, but yeah.
(Interview 3, C001)

Another, who had had cancer as a baby and lived with a long-term life-threatening condition that had resulted from his previous treatment, had begun to consider and question his mother about the implications of his condition more as he had grown older:

M013: He said, you know, "what cancer did I have?" and he started to question, like. He knew what he'd had but what… and I said… I was honest, I said, "it was called, you know, and when they stage 4," I said, "that was bad". I said, "they did say there wasn't much of a chance for you," I said "it was that bad". And he went, you know… he could understand why I panicked. And I was driving but it was a weird moment because I couldn't like hug him but he sort of thought, "oh I get it now". So he said, "I'm really lucky then aren't I?" I said, "you are, [C014], that's why I do everything I can to keep you well, you know".
(Interview 2, M013)

One of the older children, aged 17 years at the time of the interview (C002), was willing to talk about her own advance care plan and thoughts about her end of life care. She had clear ideas about being cared for in a hospice, preferably outside, with "*a special song*" playing and had discussed her funeral arrangements with her family. Despite the conversations and openness about dying amongst the family, death still seemed an abstract concept. In the example below, C002 depersonalised the conversation by referring to her uncle's death:

Mother: And what's your favourite colour? So you know if anything happens to you and you need a funeral, what colour do you want everyone to wear, even men?
Child: Pink, purple.
SM: You've thought about a lot of things
Child: Because my uncle died
Mother: And what colour was his?
Child: White and blue. But I wasn't there
Mother: No. Why wasn't you there?

Child: *Because … He died of a heart attack*
(Interview 3, M002 C002)

One child had expressed thoughts of not wanting to live anymore at a time when his condition was progressively deteriorating. His parents, struggling to manage this as well as the deterioration in his condition, described his thoughts as a "*phase*" and "*silly*". As illustrated in the quote below, it was extremely difficult for the family to discuss. The situation had been managed with a referral to a counsellor:

Mother: *He did speak to, what was it, was she a counsellor or a… someone in the team, because he kept coming out with these silly sayings, and saying horrible things about himself. And so we spoke to [consultant], and she referred him to like a counsellor, and she just come and sat with him and asked him questions and that. It was just a month or so, he was going through it, wasn't you.*
Father: *It was just a phase he went through … You don't talk like that now, do you [C009]?*
Child: *What?*
Father: *Remember you used to say silly things, like you wished you were dead, and all this kind of thing. You used to say that, didn't you?*
Child: *Yeah.*
(Interview 3, M009 F009 C009)

Focus group participants had experiences of trying to manage the unspoken awareness of death and dying amongst the children and their families. They expressed concerns that the views of the children would often remain unheard. They also provided insights into how difficult this was to navigate with the child, their family and healthcare colleagues:

Res 15: *Sometimes we never hear the voice of the child officially. We know what the voice of the child would say, if we were allowed to hear it, and its very uncomfortable. And often, erm it's the parents blocking us having that conversation. People on the outside looking in say, "you know, isn't this terrible? Because you are managing this 14 year old, not talking about the fact that he's dying, how can this be, how can you let this happen?" And it's one of the most uncomfortable things that we have to do, but at the end of the day we maintain a relationship with the whole family, and we have to sometimes just accept that you cannot let the relationship with the parents crumble to nothing by challenging in that way. And, and we know that the knowledge is already there, it's just unspoken.*
(Doctor, focus group 1)

Subtheme 2: A feeling that time may be limited
In the context of their child being at risk of serious and unpredictable life-threatening deteriorations, the families valued their time and there was a focus on living life on a day-to-day basis. Some took the opportunities to partake in memorable activities whenever possible, for example, one young person had been horse riding wearing Jimmy Choo shoes:

Mother: *She went "if they've got red sparkly Jimmy Choo flat ones can I have them?" And I went "you've got blue". "Yeah but I want red". And I said "are you going horse riding in them?""I will if I can".*

Child: *Was that at [event]?*

Mother: *[shows SM a photo] That's the best photo that is.*

Child: *Horse riding in Jimmy Choo's.*

(Interview 2, M002, C002)

Annual events such as Christmas and birthdays became particularly important, presenting for some an acute need to celebrate together or to ensure a memorable celebration and spend time together as a family:

Child: *I got out on Christmas Eve. Then came back Christmas Day.*

Mother: *Because that was my biggest thing, that all the family wouldn't be together and stuff. And they said we'll let you go home Christmas Eve, you can have Christmas night at home, or like Christmas Eve, Christmas dinner, but we want you back on Christmas afternoon. But if you spike your temperature, or anything, you've got to come back straightaway. So we did have all the family together at Christmas, and it was very emotional. But we were all together, even though we had to go back Christmas night, that was a bit weird.*

(Interview 1, C012 M012)

Other opportunities arose because of the child's condition, including invitations to charity days and family trips to places such as Legoland or Disneyland. Three of the children had taken part in photoshoots for fundraising campaigns and arts projects. One of the children had been the subject of local and national newspaper articles, and another was invited to decorate the Duchess of Cornwall's Christmas tree. For some of the children, these experiences and invitations arose so frequently that they became part of their everyday life, and they expressed some ambivalence towards them. In the example below, a child described her attendance at an event at the Houses of Parliament. Cake had been an important feature of the day:

SM: *What did you do on Monday?*

Child: *I forgot.*

Mother: *No you haven't.*

Child: *Yeah I have.*

Mother: *Where did we go then?*

Child: *London …*

Mother: *And what did we do? Did we go somewhere posh?*

Child: *It was boring… I remember cakes.*

(Interview 1, C008, M008)

Although family members appreciated these opportunities, there were times when they served as a reminder of the severity of the child's condition, as explained by M001 below:

Mother: *Yeah, so there's the… you know, and it is good and I'm grateful, but it just reminds me. DLA, Blue Badge, Supershoes. Um, we got a £170 cheque from [charity] really early on.*

SM: *And what was that for? …*

Mother: *Just to support you with parking fees, you know, they know that you're going to have additional fees, so it's all… it's good stuff, don't get me wrong and I'm not ungrateful … because not everyone is getting this sort of treatment are they, it's only because [C001]'s so ill. And I'm like, oh, okay, yeah.*
(Interview 2, M001)

7.3. Micro-system findings

Interpersonal relationships between children, their families and their healthcare professionals were highly individual. The nature of the relationships could have a profound impact on the child and family's experience of healthcare. The analysis revealed two themes relating to the importance of relationships on the child and family's experiences of the healthcare system at a micro-system (interpersonal) level as follows:

1. Connections with healthcare professionals provide a feeling of security
2. Family members recognise a need to support healthcare professionals within these relationships

Theme 1: Connections with healthcare professionals provide a feeling of security

Children and family members often described one, or a small number of individual professionals, who stood out for them as being key in the delivery of the child's care, or with whom they had an important interpersonal relationship. These were healthcare professionals who they perceived to be listening to them, responding to their needs and those of their child, and who they perceived were providing consistent support. The connections between children, families and these healthcare professionals were associated with a high level of trust, and provided a feeling of security for children and families, that was highly relevant at times when they were particularly vulnerable, such as when their child was acutely unwell. Professionals who "*came every day and brought tea, sorted the symptoms, and made us laugh*" (M002) were valued. These key trusted personal relationships developed through a variety of different experiences and perceived actions on behalf of the healthcare professionals, described as four subthemes below:

1. Children want to feel "looked after"
2. Connections become established through consistency
3. Healthcare professionals are advocates for children and families
4. Healthcare professionals being alongside children and families.

Subtheme 1: Children want to feel "looked after"
The children's relationships with individual healthcare professionals varied over time and according to whether they were at home or in hospital. At each interview, the children spoke particularly

about the professionals who they saw the most of. Healthcare professionals who spoke to them, acknowledged their needs, managed procedures such as cannulation with minimal distress, and were perceived to be "*looking after*" them (C001, Interview 1) stood out for children. As inpatients, they saw less of senior clinical staff; it was the *"kind"* ward nurses and housekeeping staff who delivered meals and were part of their daily life who were spoken about:

Child: *The nurses all sit together at the nurses' desk*
SM: *And what are the nurses like?*
Child: *Kind …*
SM: *And who else looks after you here?*
Child: *[name]*
Mother: Play lady. And [name], they're the play specialists aren't they? …
SM: *You said about the play, and the nurses, and the, who else? The teacher, who else is important? Who looks after you in hospital?*
Child points to his mother
SM: *Mummy of course*
Child: *And the dinner ladies*
SM: *The dinner ladies…*
(Interview 1, C001)

Mother: *Who do you remember from the hospital [C007], who works there?*
Child: *The one with the deep voice.*
SM: *Who's that then?*
Child: *The one who, [Housekeeping staff].*
Mother: *That's [Name], the food… well [C007] calls them the food doctors. They're the ones that…*
Child: *Serve the food.*
 (Interview 2, C007)

The children valued seeing professionals who knew them and who were able to carry out medical procedures in a particular way, for example phlebotomy with a certain type of needle that would cause them less pain and distress:

Mother: And who do you hope that's going to do the blood?
Child: *erm, [phlebotomist name]*
Mother:[phlebotomist], yes, you like [phlebotomist] to do the blood don't you. And what about the needle? What colour does it have to be?
Child: *Blue*
Mother: And what happens if it's the orange one by mistake
Child: *I don't like it. It hurts me*
Mother: See if it's a different phlebotomist that doesn't know [C008], she might pick up the orange one, ooh she's out of there, aren't you?
(Interview 1, C008)

Senior doctors and nurses were more prominent in the children's views when they were outpatients, returning to hospital and meeting these professionals during clinic appointments. The children's relationships with these medical staff members became more established as their condition became

more long term, and there was sometimes a "favourite" clinician. One child (C008) had named a teddy bear after her favourite consultant. Others developed a relationship with a clinician who had a common interest with them, such as a favourite football team.

Subtheme 2: Connections become established through consistency
Both the children and their family members valued consistency amongst the healthcare professionals providing their care. This was achieved in a variety of different ways. Some of the children and families were frequently seen in a certain department or healthcare environment, such as the Emergency Department. They were relieved to see healthcare professionals that they knew, trusting that they would know their situation and deliver the care they needed in a timely and responsive way.

Mother: It's great when you see one of the doctors or consultants that you know.
Father: Yeah.
Mother: And obviously that can't happen all the time, but you do feel relieved when it's someone that you know, because they understand him [C007].
(Interview 1, M007 F007)

In the example below, where a mother describes an attendance at the emergency department, a known healthcare professional was able to intervene to prevent delays in the care that the child received:

Mother: The lady in dark blue [in A&E} said, "oh no, you have to ring for an ambulance to go to [adult hospital]". I said "why?" She said "because she's been transitioned", I said "no she hasn't". "Well we've got alert assistant saying she's being transitioned". I said "look, we would like to be transitioned, we love the [hospital] but we need… we're not transitioned" I said, "so you need to tell us what way to go". So luckily, as I was just standing there, worried about her, because her temperature's 40 and she's rigoring, a consultant, another doctor consultant who knew her said "what's the matter [M002]?" "What's… you look upset". I said "they're saying I've got to get her"… she said "how can you? you're not transitioned?" She said now "put that young lady in that cubicle and sort her out, and I'll get Dr [Name] down".
 (Interview 1, B002, M002)

The children also valued consistency. In the example below, it was important to the child that the same healthcare professional took her blood at each clinic visit, because this particular healthcare professional was familiar and trusted with a potentially painful medical procedure:

SM: And what about, erm, is it, can I ask you is it important to see the same doctor or the same nurse? You know you said about the nurse who takes your blood, [name], is it important?
Child: [Nods head]
SM: Yes
Mother: What happens if it's not [name]? Do you mind?
Child: [Nods head]

(Interview 1, C008)

Other valued relationships developed over time between children, families and their healthcare professionals from particular teams. Sometimes the relationship became very close; one sibling explicitly described his 'relationship with a clinician as a "friendship", and a father expressed his "love" for the community nurse:

Brother: Yeah, I like the consultants. Luckily I get on with them all, because I have a good laugh with a lot of them, especially Dr [Name] and Mr [Name], they're the ones that do speak to me away from mum and dad. Don't they? Like I could see them anywhere and they'd stop and talk to me, wherever. … And Dr [Name], he was like my best friend.
(Interview 1, B002)

Father: For respite last year my community nurse, her name is [name], I like here, she's very… I love her, she's a mother, she's lovely. Yeah, lovely. She call me always, are you okay, family okay, [child] okay – yes. If I have any problems I call her, I have this problem. No problem I will see you today. She comes soon.
SM: Every day?
Father: If I need her every day she comes visit me every day. She is very, very kind of respected mother.
(Interview 1, F004)

Changes in the system, such as shift changes or a new rota that led to changes in staff availability, affected the families' experiences of the healthcare system and the continuity of care provided by healthcare professionals. Sometimes this was very abrupt and came without warning, which could leave families with a feeling of abandonment. They found this particularly difficult at times of uncertainty, when they felt their concerns and questions were not addressed, as in the example below:

Mother: Then this random doctor came up, we didn't know who he was, certainly wasn't one of the team we normally see, and couldn't answer any of my questions. So I was like "oh my god, I don't believe this". So I ended up thinking "I need answers", because now I've not been told about the X-ray, the doctors are not turning up when he's actually in pain, what's going on … I want somebody up here to explain what's going on, because my son's asking questions and I can't give him the answers because no-one's giving them to me.
(Interview 1, M011)

In the example below, the doctor who had been seeing the child on the ward went on holiday without warning the family that he would be away:

Mother: And then so Dr [consultant] was on holiday, so after he'd said right just need to get you up on your feet, and, err get the feed sorted, then, he was on holiday for three weeks. And we didn't know. So he just disappeared. He didn't say he was going on holiday, no-body had said that he was on holiday, so I was just like, what, he realised it went wrong and he just like buggered off or what (laughs)
(Interview 1, M001)

Similar experiences were described when junior doctors moved on to a new job rotation or there was a change in the team of doctors who were managing the ward patients. These changes sometimes led to professionals who did not know the family well taking on significant responsibilities such as the delivery of bad news. In the example below, the news that a disease had relapsed was given to a child and family during a weekend on-call shift:

Father: *Because they did sit down and say we've got the results, and the disease is back.*
          And then we're all sitting here, and then he said… did he say then we'll go to the other…
Mother: *That was a consultant.*
SM: *A consultant, a different consultant.*
Mother: *Because it was a Sunday, it wasn't… we've seen him once or twice I'd say, probably*
          weekends we have seen him.
Father: *We don't see him much.*
Mother: *I wouldn't know his name.*
(Interview 1, M007 F007)

Changes in staff associated with an organisational change, such as a change in the way clinics were organised, could be a significant loss. In the example below, a reorganisation of an outpatient clinic would result in the child and family's care being transferred to a new consultant after several years:

Mother: *It's devastating. It's as devastating as finding out that she [C008] wasn't going to live.*
SM: *Really? It feels that… it feels like that?*
Mother: *Yeah. … They just keep saying it's the hospital's decision. "You should go and have a*
          meeting with them", you know. I've spoken to [doctor] over the phone just before
          Christmas, and it was just a conversation that just wasn't going anywhere. Every time I
          started to say a reason that I didn't want to change, she'd have a ridiculous reason why it
          should be changed. Which mostly is so that all of the children are categorised and have a
          consultant. And we all sit in the waiting room with families of the same position. I don't
          care about who I'm sitting in the waiting room with. I want to see the best doctor that
          there is for [C008].
(Interview 1, M008)

The feeling of security that could be perceived within relationships between children, family members and healthcare professionals was easily compromised. In the example below, a healthcare professional outlines the need not only for trust within the relationship, but also for healthcare professionals to demonstrate respect for each child and family, and their situation. The example below relates particularly to discussions about the provision of palliative care and possible referral to specialist paediatric palliative care services:

R003:   *I think I would expand the word "trust", and make it, either add in or replace it with*
          "respect". Because some of the things in terms of before they meet the family, one of the key
          things is making sure they've got the appropriate information, they are the right people to be
          in that decision, you know, the discussion, and that they've made the time to find out about
          those circumstances and they're prepared to give that time with the family. But, erm, I've
          certainly seen experiences where people waltz in and assume that they know everything, and

> *actually that's very disrespectful to the family and I don't know that trust would come into it in quite the same way, but I think respect might come into it.*

(Doctor, focus group 2)

Subtheme 3: Healthcare professionals are advocates for children and families

Families described the importance of professionals who advocated for them and for their child. Actions that demonstrated to families that professionals were taking into account the child's interests and offered reassurance that their child was being treated as an individual, even in highly intense clinical situations, were also valued:

> *Mother:* *And then he went downhill the next day. And the doctors were so good. I mean they were fantastic the doctors, brilliant, all of them, really brilliant. And they'd be doing all this stuff; they'd be taking him off the oscillator to see if he's ready to move onto the ventilator and they'd be looking round at pictures he'd drawn, you know, when they got that green [drape]… I don't know what… And I'm like, don't talk to us about what we're… you just look at what you're doing, don't worry about my pictures. They were amazing … they cared; you could see, … even when they weren't on duty they used to come, they were lovely.*

(Interview 3, M007)

In the example below, a relationship between one particular healthcare professional and family had developed when that professional had taken on the child's clinical care in infancy. The child had been discharged home to die, but had survived longer than had been anticipated. The particular healthcare professional involved took a close interest in the child, and worked hard to provide life-sustaining treatments, after the child had been described by others as a *"failure to die"* case:

> *Mother:* *She [doctor] always says, this is the "failure to die" case. But she doesn't mean it in in a… she's always joked about it because she thinks it's so ridiculous that she was sent home to die. And that's actually what's on her notes, when they sat round a table to discuss [C008], she's the "failure to die" case, "what do we do with her?"*

(Interview 1, M008)

Family members valued professionals who *"really stood up for us"*, *"did everything"* and who *"used to fight my corner if something was not right"* (M002). Healthcare professionals who had knowledge of the child when they were well, and the life of the family at home (*"the whole picture"*), and who could communicate this to their colleagues, were important to children and their families. In situations such as PICU, this knowledge held by healthcare professionals could have a significant impact, including on decisions about whether or not intensive treatments were continued.

> *Mother:* *[C002] was a cheeky madam. And they just seen her keeping needing intensive care, but they didn't see [her at home], once the help, the support she needed, she goes back, they just see her leaving [ICU] still quite poorly, and not seeing the whole picture. They probably think, oh mum's just… … but when somebody else professional comes in and sees literally, yes, she's squirting me [with a water pistol] in my suit, and then she helped, she really helped try to get [C002] better…*

(Interview 2, M002)

Family members described a variety of other situations where these acts of advocacy by individual healthcare professionals were crucial. On occasions, they could be life-changing. The examples below outline occasions when significant decisions about resuscitation status and continuing treatment in critical care settings were influenced by individual clinicians who either had knowledge of the child when they were relatively well, or when they were willing to try an alternative treatment plan:

Mother: And that's how [palliative care doctor] got involved because when we came home he [intensive care doctor] was still saying she wasn't for resuscitation and we wanted her to be. So [palliative care doctor] came out and [C002] was squirting me with 50 ml syringes of water, running around, because when she was well. She was really well. And riding her bike in the house, just a typical mad day. I just said to [palliative care doctor], "I'm sorry but it is a madhouse", and she said, "you know what, this has made my mind up", she said, "where's your computer". You know when you had the big old computers, and her [C002's] dad said, "upstairs". She went up there and wrote a letter to him. He [intensive care doctor] changed his mind.
(Interview 2, M002)

Mother: It's like when [child], a few years ago, afterwards, when she was about 2, 3 something like that, she ended up really serious, she ended up on PICU. At [hospital], and they said to me that "erm, oh she's not gonna survive this", and we were like absolutely devastated, and I remember after about 2 days, when she was still on the ventilator, and this consultant Dr [name] came in, and he came in, looked at her notes, and I thought to myself, "this is another one, he's not going to do a thing", and he sort of looked at, read her notes, and he went out, and after about 5 minutes, he came in and he said "this is the medicine we are gonna give her", and he prescribed, and I think as soon as that as soon as that, the first, second lot she had, she started breathing again. And I still say to him to this day, you are the angel that saved our daughter.
(Interview 1, M003)

Other examples of advocacy included individual healthcare professionals taking a role in co-ordinating the child's care, and making efforts to ensure that particular elements of care suited the family schedule. One doctor organised clinic appointments around school times so that the child and her mother could attend. Other individuals would make themselves accessible to families, for example via a mobile phone number or email, and would provide support when the family encountered problems with a particular aspect of the child's care. A frequent example was organising repeat prescriptions from GPs in the community. In one case, a local healthcare commissioner was particularly helpful in addressing funding concerns related to the child's needs and care at home:

Mother: But we are so fortunate, our commissioner has been to the MDT's [multidisciplinary team meetings], he's been brilliant. When the hours were increased recently there was a big meeting, um, my community nurse did say, I think we will be looking at this again … and he was lovely. He'd sit there and he'd say to his team, he said, "I don't understand all of this from a medical point of view" – his background is mental health. And so he said, "I don't understand this, but if you tell me and explain why he needs it then he will have it". And I've not had a "no".

(Interview 2, M014)

Family members recognised that some of these actions may be "*over and above*" the clinician's usual job role, requiring a high level of personal commitment.

Subtheme 4: Healthcare professionals being alongside children and families
Family members noticed and appreciated healthcare professionals who spent time with them:

Mother: She would come and sit with me and tell me anything. You know, and she was always in a rush, she'd got loads to do and she'd got loads of people to see, but… She was wonderful. And she's just so kind, really, really kind.

(Interview 2, M001)

Family members expressed a desire for staff to be with them at difficult times, such as when their child was in pain, even if they could not "fix" the situation, as in the examples below:

Brother: And one of the doctors just came and sat with me for a bit and that was really important. He didn't even really talk very much, they were just there.

(Interview 1, B002 M002 C002)

Mother: He was really, like, beside himself [in pain], and I was in the room with him obviously, erm, and then the nurse sort of kept coming in, and drifting off again and coming in and drifting off again to do something, make a call, and coming in. And I was like "I'm really sorry, I don't want to be left in the room on my own with him … I'm sad, I'm scared and I feel completely helpless, can someone come and sit with me please" … I think you are pushed to a limit, you know like "No I'm sorry this is enough, somebody, come and sit, do something about this, I'm not sitting here listening to this on my own, I can't do anything about it, and it's breaking my heart"

(Interview 1, M001)

There were notable examples of when healthcare professionals did sit with families at particularly difficult times, and the impact this had:

Mother: His consultant is amazing … he's just been… he came every single morning when [C007] was unwell, every morning, he sat with us and you know that they've got other kids to see, and he sat with us… he'd sat with us because [C007] was struggling at one point, and of course who does [father] talk to? And so [consultant]'s another man and he just sat with [father] for an hour, just sitting with him, just getting upset also. I mean they're all human, they all love their kids don't they, but you need to see that as well.

(Interview 3, M007 C007)

The palliative care professionals who took part in the focus groups also recognised the impact that spending time with families, listening and not necessarily providing any other intervention could have; *"Just being alongside families in that situation is a very powerful thing"* (Nurse respondent focus group 1). The benefits of listening, and building a relationship with families which could help them to make difficult medical decisions, and the importance of having the time to do so, were described in the examples below:

Res 1:   *I think one thing, is that it's being able to listen. Actually being open to, you know, what the family, and what the child, how they're feeling, what they. Building on that relationship really. Showing that you care, and that you know,*
Res 2:   *And having the time to be able to do that as well.*
(Nurse respondents, Focus group 1)

Family members had insights into why it was difficult for healthcare professionals to spend time alongside them in pressured healthcare environments. They were perceptive to the conflicting demands placed on healthcare professionals that could compromise their ability to provide time to families, including "ticking boxes":

Mother: *I think once they go, especially in hospitals, once they come in that job its ticking boxes. See patient, after patient, after patient, and its lost that caring, the extra is lost down the line.*
(Interview 2, M003)

Theme 2: Supporting healthcare professionals within these relationships
Healthcare professionals, as well as the children and their families, valued the *trusted relationships* that developed between them:

Res 8:   *And I think that's where you get the kind of trust building up. It is a lot about that old fashioned sort of trusted relationship with the person who cares for you. And if you don't trust them it's very hard to take that advice, to take that knowledge, because a lot of what we do with children is about the knowledge of the child, not the knowledge of the disease. You know there's a lot more to it than that. And actually having that knowledge of the child and family, that gives you a much better standing to support that family going forward, than just knowing a lot about the diagnosis.*
(Doctor, focus group 4)

Healthcare professionals clearly recognised the importance of their relationships with children and their families. They described occasions where it was challenging to manage their own emotional investment in the provision of care to children and families, which caused a sense of vulnerability:

Res 6:   *I don't think it's on a subconscious level. I think it's on a conscious level. And, you know, it's one of those things that people always warn you against as a healthcare professional is getting emotionally invested with patients because of the vulnerability. But on the other side it's what allows you to have a relationship with them, and what gives you reward and what*

makes you very human. And I struggle with the dichotomy of the advice that I'm given and what feels right to me on a day to day basis …It's really really difficult because it's erm, it leaves, it does leave you with a sense of vulnerability when you emotionally invest in a family.
(Doctor, focus group 1)

This became particularly acute when there was a possibility that the child would die:

Res 5: But the most difficulty that the child has, you know the child may be going to die, family they know that, and it may involve yourself as well, think what if this is my child. They can see your expression. How do we learn how to deal with this situation?
(Doctor, focus group 2)

A need to ensure support for healthcare professionals to cope with the emotional aspects of providing care to children with life-limiting and life-threatening conditions was frequently outlined during the focus groups. It was suggested that this support should be provided in "three layers" as outlined in the quote below:

Res 15: I think for me it's three different layers, isn't it. It's the individual response that we are talking about. But having a supportive team is absolutely crucial, that you can count on, you can talk to each other. And then the third one is the actual organisational structure, working for an organisation that actually recognises that and actually puts the right resources in, structure in, to care for staff's emotional resource and resilience. So you've got to have all three. Individual emotional capacity to cope, a team that works effectively together, and an organisation that will support that as well, so I think when you've got all three, it's perfect, you've got the perfect kind of combination there. It will work with one or the other, but the perfect combination is all three layers, working together.
(AHP respondent, focus group 4)

Some of the relationships between children, family members and healthcare professionals were so well established that on occasions family members could tell when healthcare professionals had been affected by a particular event and recognised the impact that this had on them. One family described an occasion where they recognised the trauma that a consultant had experienced when another child under his care had died. They also recognised that healthcare professionals had their own family lives to manage. On one occasion, a mother had been in a position to advise her daughter's consultant to attend to his own family needs *"Doctors and nurses have lives – I said to him, you got to be with your family, we are ok here"* (M002). Another mother *(*M006) described a recognition that clinicians might be having a bad day, or have family pressures of their own to cope with:

Mother: … you've got to acknowledge that sometimes they've had a bad day as well, you know, the doctors might have had a really bad day and they might have had to, you know, sort out emergency childcare for their own child before they've come to the appointment. And you

know, if they're a bit, you know, brusque then it might be because they have had as bad a
day as you, you know, try and come from it from both sides.
(Interview 3, M006)

7.4. Summary

This first findings chapter outlines key, interrelated themes that provide insights into the lives of the participating children and their families, and hence the contexts in which palliative care must be delivered to them. The child's condition has a significant impact on family life, changing the family narrative forever. Over time, family members develop expert knowledge and expertise in the management of their child's condition and care, to the extent that the provision of this care, and management of the logistics, become a family vocation. The home becomes a healthcare system in its own right, from which family members negotiate and manage interactions with other systems. Families often have insight into the life-threatening nature of the child's condition, but the possibility that the child may die is rarely spoken about. Over time, significant life-threatening deteriorations in the child's condition, followed by recovery, become the norm.

The children who took part in this study tended to have knowledge of their conditions, but did not wish to be defined by those conditions. They expressed their own needs and priorities for life, but risked these being unheard by the adults around them.

At a micro-system level, the importance of individual relationships with healthcare professionals was a strong theme of the analysis. Relationships with healthcare professionals provided a feeling of security and being "looked after" for children and families. Connections with healthcare professionals occurred through acts of advocacy, consistency and an ability to be alongside children and families. There was recognition amongst families of the support that professionals may require within these relationships. This analysis highlights the significant impact of individual relationships with healthcare professionals on the child and family experience of healthcare, which form the foundation for the delivery of palliative care.

8. Findings 2: Meso and macro-system findings

8.1. Overview of Chapter 8

Chapter 8 is concerned with the child and family interactions with the healthcare system, including the delivery of "palliative care". The first section of the chapter outlines findings that describe the child and family interactions with individuals within healthcare at an organisational (meso-system) level. The second section provides macro-system level findings, with the macro-system being the healthcare system, and culture within that system. The themes relate to the life-limiting or life-threatening nature of the child's condition and how this is, or is not, responded to by the healthcare system, depending on organisational (meso-system) and wider cultural (macro-system) factors.

The data from the child and family interviews is triangulated with the data from the focus groups throughout the chapter; the accounts from the child and family interviews often resonated with the observations and experiences of healthcare professionals, who were then able to provide further insights into the issues that the families raised.

The findings provide evidence that addresses the research questions:

- What are the lived experiences of children with life-limiting and life-threatening conditions and their families?

- How do children with life-limiting and life-threatening conditions and their family members perceive healthcare services, and in particular "palliative care"?

- When and how does "palliative care" provide benefit for children with life-limiting and life-threatening conditions and their families?

8.2. Meso-system findings: The child and family experience of the healthcare system

The themes related to the child and family experience of the healthcare system are as follows:

1. The structure and culture of the healthcare system is based on pathological systems
2. Evidence-based guidelines and protocols create a rigid biomedical healthcare system
3. The family "fights" a fragmented healthcare system
4. Families wish for someone to "check in" with
5. Palliative care is considered a distinct specialist service.

Theme 1: The structure and culture of the healthcare system is based on pathological systems

Over time, children and their families developed detailed knowledge of the healthcare system. They from specialist and highly specialist medical and nursing teams who provided expertise and access to

the most up-to-date treatments. The parents of C007 described how they had learned, over time, to identify members of the different specialist teams in the quote below:

Father: *You did get to know different sort of teams, because there's a different sort of look about them all. You know. You tend to know it's not someone oncology when they walk in, you know it's someone from the anaesthetist's team, because of what they're wearing. So clothes often give you an idea of who they are.*
Mother: *And then we do get to know the faces. Like we would know the pain team now.*
Father: *Yeah.*
Mother: *We know the oncology doctors, the bone ones always look a bit more serious, and a bit bigger. I don't know why.*
(Interview 1, M007 F007)

The highly specialised healthcare required by the children was accessed through a healthcare system that was organised around pathological systems (organs and conditions). The focus of the work of each specialist team from the perspective of the children appeared to be one particular area. This could be a particular organ or body system, a particular stage of disease affecting that organ (for example renal teams who specialised in the management of haemodialysis, or patients who had had a renal transplant, but not both), or an episode of illness (such as a life-threatening deterioration which required admission to PICU). The children and families were aware that each specialist clinician concentrated on managing the organ system of their specialty. While they appreciated this level of specialist healthcare, there were times when they wanted a more holistic approach to the child's care:

Mother: *For a very long time it was more about "let's control, let's make the [organ] better, let's make the [organ] failure better, and everything else will fall into place". It took them a really long time for the [specialist] team to be able to stop trying to make things to do with the [organ] better … they were just focussing on the [organ] failure and saying "oh, because of her [organ] failure she's got swelling in her ankles, she's got fluid retention, because of her [organ] failure. She's vomiting because…" Well, is the vomiting because of something else that in turn is then making the [organ] failure worse? It might be nothing to do with the [organ] failure, it could be something else what's caused it. It took them a long time to stop doing that, and like getting lots of other people involved to look at everything else.*
(Interview 3, M011)

From the perspective of children and families, this highly specialised healthcare system could seem fragmented and disjointed. There were many examples of the challenges this presented for individual families in terms of managing the child's condition day-by-day. A striking example of the child and family experience of a highly specialist system is outlined in the quote below, where a mother had been provided with a Young Person's Access Card to allow rapid assessment and admission to hospital in the event of a deterioration in her child's health. Instead of one card being issued, she understood that she would be asked to carry five different cards, one for each of the

medical specialist teams who provided care for her daughter. The requirement would be that in the event of a deterioration, she would need to identify the most appropriate team for her child's admission to hospital.

Mother: And now they're telling me I'll have to carry five of them from different consultants. So I said if I'm in a panic, getting to the hospital, and I only pick one up, and it's the wrong doctor for the wrong treatment, what am I meant to do. And they said no, you'll have to carry five, so I'm waiting for the other four to come.
(Interview 2, M002)

The result for this mother, intentionally or not, was that she felt she was being asked to take responsibility for the complexity of her child's condition, having to identify the organ system that could be causing her child's deterioration, and identifying the specialist team who would be best placed to manage that particular deterioration.

In situations where a child presented with a problem that was not considered to be within the remit of a certain specialty, a referral to a different specialist team could ensue. Family members expressed some frustration about these new referrals, particularly related to the delays that the referral process caused in addressing new healthcare concerns:

Mother: you know, they can see that's not right, or test results say things, you know. Surely, you only need a little bit of guidance not a complete, "oh I can't look at that at all, I refuse completely, that's out of my… [specialty]". That's ridiculous. The actual… "the GP's for coughs and colds", you wouldn't see a different GP… "Oh sorry that GP doesn't look at throats, you've got to go and see that one for a foot". Like we have dentists and doctors it would be like, "no we'll have doctors for legs, doctors for arms". It's just insane.
(Interview 3, M008)

There were also frustrations about the practical challenges that referrals to new services could cause, including a need to co-ordinate multiple clinic appointments:

Mother: Because all of the departments are independent so they don't know, but they should be able to go into the system for that patient and see where they've got other appointments and say, well okay, we'll slot them in there and that works better. Not like on a separate day, it's just mad.
(Interview 3, M008)

Two further examples described by both families and healthcare professionals as areas of concern were admissions to intensive care and referrals to specialist teams for pain management. Admission to PICU required children and families to meet PICU staff and engage with the clinical team. They quickly had to build new relationships with clinicians, particularly if their usual care team withdrew at the time of admission, effectively delegating responsibility for the management of the acute

episode to the intensive care staff and leaving the child and family to negotiate their new relationships:

Res 6:   Then, they hit the transition of, OK, they're acutely unwell now, they are life-threatened acutely, And quite often we see that specialist nurse, or that person who has been a support, now, they'll feel it's inappropriate to come at that point because, and I don't know whether that's their own, erm, coping strategy, but you certainly get that in that transition [to intensive care]. So you know they trust that person, but that person's not there, and now they've got to engage with a whole new team, or a whole new person.
(Nurse, focus group 4)

Pain management was often deferred to the specialist pain team. This had the benefit of obtaining specialist advice and expertise, but could also result in delays in pain assessment and the administration of medication for pain relief, which was distressing for the children and their families:

Mother: I mean we don't obviously know what it's like to be a nurse on this ward, and I'm sure it's very busy. But things like bleeping the pain team, to me doesn't seem a big task. So when that hasn't been done for three hours after it being asked to do, especially for him.
(Interview 1, M007)

Mother: And because he'd got his pain relief pump, he'd been doing this all night with the button. So he'd actually wore himself out. And she come and she looked at him, and she went "you want pain…, oh, he's resting, he's sleeping". And I went "oh…" well I was actually shocked, that's because he's been pressing that blinking button all night, and he's worn himself out. Oh well she says, I'll leave him then, and just refer him to the team that are on today. And I was like, well if you'd have come earlier, you'd have seen the pain that he was in. Now, you know, you've not seen it. So I was very narked at that point.
(Interview 1, C012 M012)

Receiving a series of onward referrals to new specialist teams led to further challenges in the family's relationships with the healthcare system. The more specialists that were involved, the more difficult it became to manage the system. As well as these practical challenges, such as arranging appointments at similar times, the children and their families had to develop an understanding of the different specialist opinions they received in relation to particular aspects of the condition, which could conflict with one another, as described below:

Child: A bit annoying sometimes because there's so many appointments to go to and it's all different people.
SM: And they're all on different days as well are they?
Child: Yeah.
SM: And do they all have the sort of same idea about your health or do you find that their ideas are a bit different? Do you have to do a lot of work together to pull it all together?
Child: They're all different.
Mother: It's different because it's for each individual different problem isn't it?
Child: Yeah.
(Interview 2, M013 C013)

Despite the sometimes vast numbers of professionals involved in the child's care, families could still feel unsupported at critical times:

Mother: To be honest, if I really sit and I think about it, there isn't any services that's responsive, there isn't any services that will actually work to benefit, it's literally me and my husband. In total we've probably got about 40 people involved with her care at that moment. When it came that [need for improved symptom control], it was literally me and my husband fighting.
(Interview 3, M003)

Opportunities to see clinicians from different specialities at the same time were rare, but valued by children and families:

Mother: We were stressed, he was getting upset, and to get all the consultants in that room, that worked.
SM (to C013):   Were you there as well in that meeting?
Child: Yeah.
SM: How was that?
Child: It was good because we had all the doctors there and like talked about what's going on and everything.
SM: Yeah, and did you feel… because that's quite… they were obviously all people you'd met before, but did they listen to what you have to had to say?
Child: Yeah.
(Interview 2, C013 M013)

The family experience of this fragmented system resonated with healthcare professionals. Focus group participants described the difficulty that families faced managing the many different speciality teams involved in their child's care.

Res 7:   Sometimes there are so many people involved … it's difficult. You know, everyone's got a role to play but for these families they've got so many professionals, it's about who's role, who's doing what … but I think, for these families there's so many people involved, it's quite overwhelming.
(nurse, focus group 1)

Inconsistent, unreliable communication between specialists both within one clinical setting (the hospital), and between different clinical settings (community and hospital), was a significant concern. Families described the challenge of ensuring that the correct, up-to-date information about their child's condition and care was shared appropriately, and were "*thankful*" when this happened:

Mother: So yeah, [Name] now is under the same, Mr [Name] for legs, and Mr [Name] for his back. So yeah, they do talk to each other thankfully.
(Interview 1, M006)

They could not assume that other healthcare teams were aware of changes in the child's management plan. They described losing contact with particular teams, depending on the current management plan for their child. For example, when their child's medical management was primarily organised by hospital teams, they would have very little contact with community healthcare professionals:

Mother: *We know just to take him back in if we're not sure. His community nurse is devastated because she hasn't seen him all year, his community nurse, because he has all his bloods and everything done. She said she might just call round at some point. Yeah, and he misses her too.*
(Interview 3, M007)

Communication between hospital and community teams seemed to be lacking, as did understanding of services in each setting. In the example below, community care was promised by hospital teams, but subsequently didn't materialise:

Mother: *There was nobody linking. At the time, at the time initially of discharge … We were told we'd have a paediatrician who will come and pull everything together, I remember that one, but I didn't have anyone, like*
SM: *Do you have a community paediatrician now?*
Mother: *No. So I dunno whether that was because we got missed somewhere, or whether it's just not, it's something that's not, wasn't done, so like, cos initially they were like oh you'll have a paediatrician, don't worry, they'll speak to everyone on your behalf and get everyone , to liaise with each other. If you've got to go to hospital for two different appointments, make sure they are on the same day as each other, same week, make sure they are on the same day, stuff like that. Never did, yeah*
(Interview 1, M005 C005)

There were also concerns about liaison and communication with primary care teams, which had a direct impact on the child's care. One mother described the GP as "*realistically, the core of it all*" (M003), but another reported that she didn't *"have the strength to battle the GP"* (M005). GPs were not always willing to prescribe the child's medications. Sometimes this was explained to the families as the medications being highly specialist, but on occasions, GPs declined to prescribe any of the child's medications, without any clear rationale being evident to the families:

Mother: *Two years ago when all this changed with [C002], three years ago, the GP was still doing [C002]'s prescriptions, all the medications, everything. Then 2 ½ years ago refused to give any of [C002]'s medications. We get nothing off the GP now. The [name] pharmacy and the children's hospital are doing it now. We're having a right struggle with them. … three weeks ago I rang up the GP because a nurse at the hospital said that they'd given me all the correct information, the size of the patches, everything, it's fine [C002], and then, they said no, it's not on the [formulary]" … Refused to do them, again. So then we had to go to the children's hospital for [consultant surgeon] to prescribe us all our creams, so we had to go all the way*

back to the children's hospital to get cream, cavilon. … It was cavilon sticks, it was mefix tape, and erm mepore dressing, and a silver dressing, and wouldn't prescribe anything.
(Interview 1, M002 C002)

This caused significant anxiety:

Mother: The medication is a part of keeping [C002] well, so I think that's why I worried about it … And that's why I think… because my husband was… he's been worried about it because I thought, oh it's just me being mum, worrying… lying in bed worrying about it. And he said, no he is because how are you meant to look after her and know… we didn't know where the [medication] was coming from or anything. And that's her life, that's what keeps her here. So we were worried about it all.
(Interview 2, M002)

Families struggled to maintain up-to-date knowledge of all of the services that were available. "*We can't ask for it if we don't know it's there*" (M006) was a frequent theme. Adapting to service changes provoked a range of antagonistic responses amongst family members, including confusion, anger and frustration. Some expressed a feeling of resignation to the news of another system change. Families also described the impact of the reorganisation of a service or the introduction of new services, designed to address a certain element of the child's care, as a further cause of disruption, particularly if these changes or new services were introduced without consideration of the child and family's individual circumstances. One example was a new service to help with transition to adult services, which was introduced to a child and her family after several months of organising their transition with their usual paediatric consultant. The new professionals were regarded with suspicion, particularly when the extra advice that they provided was complicated and did not seem to fit with the child's circumstances. Both the relationship with the new professional and their input into the child's care required careful negotiation:

Mother: Do you really want to know the truth? …Because I don't drink, but I think I need a gallon of wine. … It's driving me insane. I don't know how parents… ours [transition] was going really well. Dr [consultant] had sorted everybody out, everything was going on track. Then we went to clinic and a lady turned up who said she was the new transition worker.
(Interview 3, M002)

Healthcare professionals equally felt that the re-organisation of services was challenging, particularly when it was perceived as having been imposed and likely to compromise the continuity of care that they could deliver:

Res 4: … we do often know children over many years and we probably, the accessibility bit is a bit challenged because we're very constrained with our volume of work, but if there's a particular piece of work to be done it might be that some of these other people take on a more intense piece of work, but I think we do provide the continuity if our employers allow us

to stay working in one area with the same caseload and don't re-organise us too often, which
is the challenge.
(Doctor, focus group 2)

Healthcare professionals described feeling that the importance of relationships with children and families, and relational continuity of care, was underestimated in the planning of system changes, or when services involved in the child's care changed. This was particularly pertinent for the organisation of care for children with life-limiting and life-threatening conditions, where there was a possibility of dying. In the example below, respondents reflect on the impact of system changes on relationships between healthcare professionals and families, and also on the change in services for the family after bereavement:

Res 11: I don't think we acknowledge the impact that that has had on the family. It may not, may not
be the child, I think invariably it would be the parents or the extended family and I think we
have to acknowledge how important that can be and how actually at times how devastating
that can be. And, you know, if that's, that's the fact that there's the death of the child, so it's
the end of life, do, do many of us see them afterwards? We might see them at the funeral,
we might see them once more after that, but actually invariably I would imagine that's not
enough.
Res 6: It's a really vulnerable period where they need it most, everyone kind of just (indicates
leaves).
(Nurse respondents, focus group 4).

Theme 2: Evidence-based guidelines and protocols create a rigid biomedical healthcare system

As well as the practical difficulties associated with navigating a fragmented healthcare system, children and their families described experiencing a biomedically driven, cure-focussed, evidence based system where concerns were responded to through the provision of a clinical or medical solution, such as a new drug treatment or an onward referral; somebody doing something to try to "fix" the problem. Healthcare delivery driven by clinical standards, protocols and guidelines, that provide clinicians with a structure for their medical decision-making, did not always seem to acknowledge the complexity that the children were living with, nor the uncertainty that they faced day to day.

Families were perceptive to the occasions where guidelines seemed to be rigidly applied to their healthcare. They described this approach as sometimes lacking "*common sense*". They were keen to know when clinicians were using guidelines or protocols, so that they could effectively negotiate and plan their child's care:

Mother: No, they never clarify following a protocol or guidelines. The [clinician] said after a while, it
was NHS guidelines. And I was like "I know", but sometimes common sense should be

more… you get something in a paper, you're not going to follow that to a tee, because every child is different. Every child's needs are different. That is just a guide, it's not written that it's a definite, if you don't do it you're going to get sued, or something's going to happen to you. It's a guideline. And then they're like "Oh, okay. I should have clarified that for you". And they're quite shocked because I'm clarifying, and I know that much, that it's a guideline

(Interview 2 M003 F003)

Mother: … we've gone from every two to three weeks, to eight weeks [between appointments].
SM: Oh right. That's quite a big change.
Mother: So I said "oh my word, what's that going to be like?. "Well, protocol is actually three months". "Really, okay. Sorry about that, but [doctor] sees her every two to three weeks" because you fluctuate so much don't you, with your bloods. But hey, protocol says. So I don't know what sort of mess you're going to be in.

(Interview 3 M008 C008 B008)

Families described occasions when they felt that their child's condition was not managed well because they did not fit a clinical guideline of protocol:

Mother: Sometimes if she went to the GP surgery and they looked at her, "there doesn't seem anything wrong". He would have to take tests. So, I think it's just about that, them people listening to us, and saying, it's like we know the normal signs of a urine infection are the urine, sometimes there's a temperature, things like that. But with [C011] it's not always the case. There's other signs that come. So we know like, she'll stop, her eating will start to decrease. She'll become very lethargic, sleeping a lot. We notice them signs, where a doctor can't see that. But it's about them listening to us and just taking our word, because we know her, and we've managed the condition for such a long time, it's important for them to just listen to us. Like when we took her to [hospital]. And we tried to tell them that she'd got a [infection]… but they still wouldn't have it, would they.

(Interview 3, M011)

On some occasions, the application of guidelines and protocols were a dangerous barrier to children receiving the care they needed, in this case urgent medical care for sepsis:

Mother: We got to this hospital and the lady was there saying, I don't know what, "sepsis, what do you mean sepsis?" I said "No, you need to get me a nurse now" And she was like "No, no, no, we've got a protocol that you have to go through." I said "If you don't get me a nurse, I'm walking in there and getting a nurse" … So she said "okay". So a nurse came out and she took one look at [child] and rushed her into resus. And she said "how long have you been there?" I said "five minutes". She went "What?!", I said "yeah". So all the doctors came rushing in, taking bloods from the lines and everything ..

(Interview 3, M002)

The rigid application of guidelines or protocols left the children and their families feeling that their needs and underlying concerns were ignored, unheard or unaddressed. Even when flexibility around a guideline was applied by one healthcare professional or team, they experienced problems in other parts of the system. In the example below, a medication for epilepsy had been carefully titrated to suit the child, but nursing staff at a centre for respite care had difficulty accepting this:

Mother: And he [consultant] was very much, well you just gradually increase and stop when you think you're there. Which was great for us, not so great for people like school nurses and respite nurses who want to have things written on exactly, you know, and they don't… they're not happy with it being, well this is what the parents decided that their child should be on, you know, it has to be signed by the consultant. And it was quite difficult that… I think [C006] went to respite once and they said, well "she should be on this dose, the protocol we've got says she be on this dose by now". And I said, "no, we decided to stop at an earlier dose". "But you can't". "We can, we were given permission to do that". And it's like, "we have to have written permission to do that". It was just a nightmare. And it's about all the people trusting and perhaps the consultant who had made that decision to let you have the choice putting in writing somewhere that we've let the parents make the choice.

(Interview 3, M006)

Focus group participants had witnessed these experiences. They described occasions where a guideline or protocol-based approach from clinicians was interpreted as the child being cared for "*as a disease, not a person*" (nurse, focus group 4). The approach that was required of clinicians to manage the clinical challenges presented by the complexity of the children's conditions was summarised by one participant below:

Res 8: These families become specialists in their diagnosis, their disease, their disorder, and actually for a lot of, certainly for my workload, they aren't standard practice, they aren't standard guidelines, they don't fit standard BNFC directions, so actually the people they trust tend to be the ones who will listen and go away and look at that, and come back and reflect and say actually this is an option. Because again in hospital particularly if they are seeing a junior doctor, they may not see their standard consultant, what they get it, the standard practice. And for a lot of these children, standard practice doesn't fit does it?

(Doctor, focus group 8)

Theme 3: The family "fights" a fragmented healthcare system

A parallel process of learning and adaptation relating to the healthcare system accompanied the continual process of family learning and adaptation related to living with the child's condition described in Chapter 7. Family members were required to become experts in navigation of the healthcare system, which was fragmented, rigid and sometimes failed to acknowledge the uncertainty with which the children and families lived. This was difficult and could be all-consuming. "*Everything's a fight*" was a common statement throughout the interviews; every interview transcript contained data about the "fight" that families had to take on in order to access the care that their children needed. They described doing whatever was needed to manage this fight for care:

Mother: It took a lot. Because I remember in the beginning it was a mission, we were having so much trouble, and the receptionists we were having to call erm, I had to call a major meeting, and erm I had to put it on the table and say well this is how it's going to be from now on, if it's not then I will just complain. Do whatever I need to do.

(Interview 1, M003)

The families, who were already in a highly vulnerable position because of their child being seriously unwell, were forced to adopt a proactive, assertive approach in order to access the care, equipment and medication that their children required. This led to a range of antagonistic responses from family members, particularly if the care needs they were trying to obtain were considered essential, such as a wheelchair or regular repeat prescriptions:

Mother: You know they do what they need to do and then that's it, it stops, unless I miss something and then I have to phone the secretary up .. It, it really does my head in, gives me a migraine … I just go for it, I have to … I've learned from experience that you really have to put yourself out there, if you're going to sit at home and think oh they're gonna give it to me, you know bring it to me, it doesn't happen. And I know a lot of families who don't get what they need let alone what they want because they're waiting, they're being told "oh it's not available, oh we'll get back to you", and it never happens, and they really struggle, they're at crisis point … every day is a struggle.
(Interview 1, M003)

Families described difficulties managing other circumstances such as changes in the child's condition precipitating a transfer to another care setting, including discharge from hospital into the community. Organisational and system factors, including poor communication across care settings, added to the fragmentation of the care that the children and families received. Arrangements for out-of-hours services both in hospital and in the community were a particular cause of concern. Family members described difficulties in being able to access responsive healthcare services out-of-hours:

Mother: And we were discharged on a Friday. That's happened a lot of times, we get discharged on a Friday and it will be, like, a bank holiday Monday, and lots of times it's happened the doctors don't give you enough stuff [medication] to last you til, til er, Tuesday at least. So I call the bank holiday Monday to the local nurses and they put their phone, and at that time they didn't even know [C010], and ask them for some help for er some and they say you are not under our care so we cannot help you. The next day a different community nurse come and I was quite angry, I say erm whoever called me on Friday that's what I had been told, if you need some help let us know, and on Monday they said we cannot help you.
(Interview 1, M010)

The possibility of seeing a professional who knew them affected how and when the families would choose to access healthcare. Some families chose to wait until they knew that a clinician who was familiar with their care would be present before accessing emergency healthcare:

Mother: But things don't happen at the weekend, and you're thinking I can't wait for Monday so we can see someone that we know.
Father: You wouldn't want your child to be ill of a weekend in here.
(Interview 1, M007 F007)

Res 3:   I know of quite a few families, if their young person is unwell at home, they will phone up the assessment unit, find out which doctor is on call, erm in the assessment unit and based on that information they will decide whether or not to bring that young person in. Erm and that's because they know that there are some who know their young person, know how to manage their young person, whereas others they end up not doing the way, treating their child the way that they would want their child treated. And it's very very interesting.

(Doctor, focus group 4)

Healthcare professionals recognised and acknowledged the concerns of children and families, and the daily fights that they described. In the example below, a doctor expressed a desire for healthcare provision to be less of a "battle" for families.

Res 3:   And although we are talking about their complex needs, actually for the families, their journey, their path to get things in order in to be able to help their child, that is what needs to be made simple, because the amount of battles these families have just to get one simple thing done, and it shouldn't have to be a battle, they have a battle every single day of their lives, just to keep their family going, just to keep their child going. Everything else should be simple.

(Doctor, focus group 4)

Theme 4: Families wish for someone to "check in" with them

There was a desire amongst both family members and the children for a clinician who would regularly "check in" with them. While they were at home, contacts with healthcare professionals could be sporadic and often in response to a new problem, rather than as a result of a more proactive approach:

Mother: Even at the time when we used to have them, I kind of had to, I felt that I was constantly chasing everyone around, like, the health visitor, if I didn't contact her she wouldn't contact me sort of thing, like, yeah, so it's not like, like you know phoning you up and how are you? How are things going? Knowing that … actually having someone around and stuff

SM: Do you think that would make a difference

Mother: Definitely, I think that it would have been more helpful for me in the sense of that obviously we knew there was support out there. There was none of that.

(Interview 1, C005 M005)

One of the children highlighted the potential value of having a healthcare professional who "checked in" with him every so often. He felt that this was missing from his care, and explained that he would feel reassured if it happened:

SM: From your point of view do you think there's anything that would improve things?

Child: Um, probably like a daily… well not a daily but like monthly sort of phone call to see how you're doing and everything.

SM: Just to check in?

Child: Yeah, just to see how you're doing, because … there's space between the appointments so they don't know what's going on in between.
(Interview 2, C013 M013)

A number of parents shared his view:

Mother: I said that to [Name] who is in charge, I said that to her, that it's really important. No matter how confident a parent is, you know, we still need… I mean I still need to have, I still need someone to fall back on, I still need that reassurance, you know.
SM: Yeah.
Mother: I do have my moments.
(Interview 2, M003 F003)

Where there had been a longstanding relationship with a specialty team, it was felt that maintaining that relationship, even if the child transferred to the care of a community and / or palliative care team, would help children and families:

Res 8: because they don't, they know, although they acknowledge that the specialist can't really do anything, but they still might just keep checking in.
(Doctor, focus group 4)

It did not necessarily have to be a specialist who would check in with a child and family. One mother described the value of having a GP who had known her for many years and who therefore knew about her two previous child bereavements. She used opportunities such as her child's chronic disease reviews to keep her GP up-to-date with how he was, and ensured that the GP saw her child when he was well:

Mother: I think we're quite lucky that I can talk to the GP, but she is a GP that we've had since before I had children, so she does know the back story and she does know all the history. But I know lots of people don't have that.
(Interview 3, M006)

Theme 5: Palliative care is considered a distinct, specialist service
Each of the families who received palliative care services considered "palliative care" to be a distinct, specialist service. One respondent described the role of a paediatric palliative care doctor as follows:

Res 14: A lot of my job is symptom control and accessing services, and that I'll interface with families and walk alongside them for a bit, and then I might pass them back to their main consultant for a bit, and then may get involved a bit later. And so parents then get used to the fact that sometimes I'll be quite intense and see them every couple of days, for a week or two, and then I might step back for, you know, six months, a year of whatever, and then come back, be involved again, and then as you were saying [doctor], it becomes a bit OK. And so when it's a crisis time, it's not odd that I just kind of parachute in, and sort some crisis out, and then come back out again, because you know there's a lot of children and otherwise you'd

get completely overwhelmed with children. So I don't "own" many children, but I might interface erm…

(Doctor, focus group 4)

In reality, the role of palliative care services varied. These services provided a particular aspect of the child's care, ranging from complex symptom control to respite care at the hospice. For the children in the study who had experience of palliative care services, referrals had been made by a variety of different professionals and had occurred at different time points in the child's illness. For one family, referral had been made at a time when their child was critically unwell in intensive care and was thought to be dying. For another, a referral was made because their child was their second child with a particular life-limiting congenital condition. Children's community nurses had referred two of the families to their local children's hospices for respite care. For one family, the palliative care team were introduced when there were symptom management concerns. This mother described learning that her child had been referred to the palliative care team only after they had been introduced for symptom control:

Mother: It's mostly like for pain. They were involved since he had like that [problem] …
SM: Oh yeah, you said, sorry. And when the nurses here said that they were coming, did they give the name of the team?
Mother: Yeah
SM: Yeah, so have you heard that word, palliative care team?
Mother: [shakes head] I didn't know, I just. I didn't know, it was that it was called that, and then I found out that that's what they do … Mmm, the first time we meet them when, when he had a [problem], the doctors say that they come just to assess his pain

(Interview 1, M010)

For another family, a referral had been made to the palliative care team because they were the only community nursing team willing to administer a certain monthly injection:

Mother: he was having injections in either leg, the doctors did get me a thing called the [name of service] palliative care team, and they come out and helped do the trial otherwise I was having to go up the hospital, which that was like massive. It was massive just having that is like… I don't know what I'd do without them. If they would take away like that sort of care… he has palliative care now once a month, they come out and administer his [injection].

(Interview 1, M013)

The themes of fragmentation and rigidity in the healthcare system extended to palliative care being a distinct and separate service; referrals to palliative care services resulted in the introduction of more professionals into the child's care, and were also frequently defined by referral criteria or protocols which would define whether the child and family could access the service or not. In the

example below, the children and families had not been able to access hospice services, or fit specific criteria to receive ongoing care from them as their child's condition fluctuated:

> Mother: You see nobody's ever offered hospice respite for either of mine, even when they've had… you know, they have quite severe [condition] and [C006] has [condition] whereby he could fall into a coma and stop breathing…

(Interview 2, M006)

> Mother: Yeah, because… because they [the palliative care team] was going to let us go. She didn't fit the criteria. But then when she got this poorly this time, she fit the criteria again.
>
> SM: Okay
>
> Mother: Yeah. So you know when she picks up, well she is picking up again. So when she picks up again they'll probably say "no" again.

(Interview 3, M002)

The professionals who took part in the focus groups were all involved in the delivery of palliative care to children in some way. Not all were members of specialist paediatric palliative care teams. They recognised this tension between meeting patient need and providing a service within a finite resource. The participant who provided the quote below was clearly uncomfortable with the idea of declining referrals, but described the need to balance this with the limited resource available for the specialist service:

> Res 1: We've kind of refined this really annually, and we've started to refine it more, because the cohort of children surviving with complex illness is getting bigger and bigger. And so we decided that we would have er referral criteria … So we have a discussion about "is this an appropriate child? Does this fit our referral criteria?" Erm we tried to make sure that our service is provided to those that need it most because we don't have a bottomless pit. … We do decline, we do decline referrals, and we will gather more information if we need to, erm so we have, we have a good system for referrals I think, mostly.

(Doctor, focus group 3)

Focus group participants described important aspects of the care they provided. Firstly, they recognised the complexity and fragility that the children and families lived with. They described the importance of respect for this situation and the individual needs of the child and family, placing their priorities firmly at the centre of the care they delivered. There was an emphasis on holistic care and the professionals recognised the importance of listening as an intervention:

> Res 1: I think one thing, is that it's being able to listen. Actually being open to, you know, what the family, and what the child, how they're feeling, what they. Building on that relationship really.
>
> Res 1: Showing that you care, and that you know,
>
> Res 2: And having the time to be able to do that as well. Staff and resources
>
> Res 3: Time

This approach to care was not described as being specific to the specialty of palliative care, but was considered as "powerful" and valuable to families, mirroring the themes from the child and family interviews regarding key personal relationships with professionals:

Res 12: *You just feel like you can do, just make a little bit of a difference. You know, it might be something very small, it might be that you can't change the syringe driver, but you can sit, and you can, I don't know, play with the child, or you know, talk to the dad, or a grandparent. Or just try and make things a little, I can't, I won't use the word "better", erm, but, give them some quality time, and be there really.*
(Nurse, focus group 1)

Res 14: *I suppose the other thing is being alongside people, even if you can't make their disease ultimately better, you can be with them and make what's left of their life as good as it can be. And that's probably why a lot of us do this, as well, I would say. Just being alongside families in that situation is a very powerful thing.*
(Nurse, focus group 1)

System concerns, such as a lack of time for professionals to spend with children and families, and reorganisation of services that caused breaks in continuity of care, were highlighted as barriers to the delivery of this approach to care by non-specialist colleagues. It was also recognised that in current policy, where there is a focus on tangible, measurable outcomes, the impact of these elements of care could be overlooked.

Res 11: *The value we add is very hard to measure, it is, and it's going, it varies from one family to the next about what they take from that relationship, or service. And that in itself is inherently difficult to quantify. You can tick the boxes about where they died, or if they had a care plan, but actually what does that mean? What about all the work to get to there, to achieve that.*
(Nurse, focus group 1)

Res 1: *So how do you measure the fact that you know, you have, you have managed over time to make an informed decision, about ceilings of care, through a series of multiple conversations, through having kind of careful discussions, through them knowing that actually you know you've reframed them, and that has then enabled them to have a functional relationship with their partner, you know, for them to get back to work, that's just not measurable, but that's a huge amount of what this team will do.*
(Doctor, focus group 3)

A clear theme from all of the focus groups was the desire to improve palliative care delivery for children and their families. However, there were diverse views about the best way to do this. The specialisation of paediatric palliative care was an area of debate. Some focus group participants outlined a clear need to develop the specialty, and the evidence base to support it. They raised significant concerns relating to professional expertise in symptom control at the end of life:

Res 1: So if they don't know, they won't necessarily, they think they're doing it, or they think they're delivering it already, or they think it's all covered

Res 2: They think writing up morphine and midazolam is doing palliative care, and the patient dies, but they don't die in pain.

Res 1: Yeah, so we have a slightly different perspective on that, erm

(Doctor respondents, focus group 3)

Respondents highlighted a need to increase training opportunities for all professionals and increase effective collaborative working:

Res 2: We need investment. I'd like a managed clinical network, lots of funding, I'd like more training places at the Royal College, because we've got a limited number and we've got loads of places that want to appoint consultants, erm I'd like to have more evidence as [nurse] says, I'd like to be able to recognise the intensity of this work. I'd like our team to have time off to do research … We don't get the opportunity, to you know to learn and develop as much as we should be … But probably about, I've been doing it for 20 years, and God it's the same kind of same people. We've had very little, we've only really been trying to get off this, you know, cycle of trying to get more and more people involved, and really trying hard, in the last 5 years to do that, but it's, we need, we need more momentum, we need money to do that.

Res 4: And nursing needs to be looked in to, because I think in the future there won't be as many nurses in the future, and I think this whole field at the moment has got a whole team of people who will not be around in another 10, 15 years, and I'm not just talking about [hospital], I'm talking about in the community, in palliative care, erm. I think. I'm not sure we've really invested in nursing, in training nurses in this specialty, and … so I do worry about this in the future.

(Doctor and nurse respondents, focus group 3)

There was tension between this desire to develop the specialty, and the potential "over-specialisation" of palliative care for children, with a need for the wider workforce to be able to deliver palliative care described. This was based on an assumption that clinicians possessed many of the skills required to deliver palliative care despite not being a palliative care specialist:

R001: Can I say one more thing. I think there's a real risk of over-specialising palliative care. You know, a lot of it is not rocket science, it is good medical, nursing, AHP care, and I think there's a risk in us. And we try and fight against it all the time in the hospital, this isn't care that has to be delivered by one of us four professionals. And I think there's, there's something about, people have those skills already, it's not making it difficult by labelling it.

(Doctor, focus group 2)

A need to ensure that services could meet the complex needs of the children was suggested as a priority:

Res 1: Oh that's another one on our wish list! We just want the hospices to do be able to do what it says on the tin. That's all we want … We want them to be able to do IVs,

154

Res 3:   *And that's very organisation based, you've got some who do respite, some who do end of life, some that won't*

Res 2:   *And hospices that will invest in medical cover. Not expect us to provide it [for free]. So one of the hospices that is having a new building, that's really cool isn't it, they've decided they won't do respite, they are going to do complex symptom management and end of life care, and I said to them, "oh that's interesting, where is your medical cover going to come from" and they said "oh that's where we need to speak to you". Yet another hospice that thinks they can throw a little bit of money at us, to do like one session a week to manage patients who are an hour and a half away and very complex, we can't do it. But they're already fundraising on this premise, and they haven't even spoken to us about it.*

(Doctor and nurse respondents, focus group 3)

In terms of improving access to specialist paediatric palliative care services, focus group participants described several examples of effective strategies. Working closely alongside other clinical teams, forming integrated teams, was one successful strategy, but was variable between regions and teams:

Res 3:   *Your integration with different teams varies doesn't it? So for example in PICU you're quite well integrated, but maybe for fetal medicine, or renal there's less in-roads at such an early stage. But we need to be spread across all of those.*

(Doctor, focus group 2)

Res 16:   *We have in-reach, so we have the hospices coming in to our NICU now in a weekly basis, so they become sort of part of the team. And it's much easier for them to be introduced as part of the team, and sort of parallel planning before you get to the stage where you're desperate for a referral to palliative care services. And getting in there as early as you can really.*

(Nurse, focus group 1)

One participant described a change in service delivery such that hospice services were introduced to children and families as a routine part of their care, which seemed to be effective:

Res 3:   *We've now taken a stance that actually for the vast majority this is just routine, this is part of your support package, erm rather than waiting until actually they've relapsed, and ah they've relapsed again, and actually they now are for end of life services. So we decided to take the much earlier stance of, this is just normal,*

SM: *Do you name it palliative?*

Res 3:   *Erm, yep, we say we'll be referring you to the [hospice], so that you can get some extra support, and your community nurses are part of the complex and palliative care nursing team, and erm but we always emphasise it's about the additional support for your child and the rest of your family.*

(Doctor, focus group 4)

8.3. Macro-system findings: Uncertainty, the "collusion of immortality" and palliative care

The families experienced a healthcare system that was fragmented, but also rigid, and there was often a lack of security provided in the form of an identified professional checking in. Underlying, unacknowledged uncertainty about the child's condition, and what might happen, further complicated the situation. The macro-system findings relating the child and family's experiences and perceptions of the culture of the healthcare system, including perceptions of palliative care, were divided into three themes, as follows:

1. The unspoken background of uncertainty
2. The "collusion of immortality"
3. The "p word" problem

Theme 1: The unspoken background of uncertainty

The children and their families lived with huge uncertainty. As described in Chapter 7 (section 7.2), the children's conditions were associated with unpredictable, fluctuating disease trajectories. As well as this, their problems and symptoms did not always have a biomedical explanation and could not always be effectively treated with an intervention. Furthermore, their conditions developed and changed as they received different medical treatments, many of which were at the forefront of medicine, for example drugs from a new drug trial, or innovative and individualised surgery. The uncertainty that this created was not always acknowledged. The healthcare system, that was evidence based and guideline driven, sought certainty. It was felt that this cure-orientated, solution-focussed approach may be even more prominent in the care of children than for other patient groups:

Res 23: I think there's something about paediatrics. Going into paediatrics, most of the time, you do fix children, and they get better, and I think that's something that on the ward the majority of us couldn't deal with children dying because that's not what why, they get better, and I think that's the difficulty
(nurse, focus group 2)

The effect that this had on the experience of children and their family members was multiple referrals to new specialists, seeking an answer, an explanation and certainty about what was causing the problem, and some kind of medical treatment. When their clinical concerns did not have a straightforward answer, and their clinical teams did not openly acknowledge this, their own thoughts that their problems may not have a medical solution were not validated. In the example below, C013 and his mother had the opportunity to discuss their concerns with all of their specialty consultants in one meeting. The meeting had been preceded by a long period of time when they had

been managing conflicting advice about how to manage an unrelenting symptom. Bringing the specialists together allowed for the uncertainty to be openly acknowledged for the first time:

Mother: I think they were shocked … it's upsetting because we got to voice how stressful it is. And I said, "not one of you have said what's causing [the symptom], or it could be everything that's causing it, or, you know, not one… not anyone has made a point" … obviously I'm asking all these questions and they all agreed that it's all of his [conditions] isn't it… obviously they said to you didn't they [C013], "you do understand that you've got all these problems?" He said, "yes". "And that you're always going to be in hospital, but we are going to look after you". … "but there's nothing we can do for your [symptom], it is probably everything" so that's the first time they acknowledged, yeah, it's a mixture of all of it and that makes your [symptom] worse.
(Interview 2, M013 C013)

Focus group participants acknowledged how difficult it could be for healthcare professionals to acknowledge uncertainty in relation to a child's condition. They described the emotional conflicts that could arise for individual clinicians, particularly when the child's condition was life-limiting and the future was very unpredictable:

Res 5:   He [a patient] feels everything for him is ok, but, you never know. But you have conflicts inside your mind. What am I doing? What should I do? I don't know.
(Doctor, focus group 1)

They described a desire amongst professionals to "listen to fix". This was possible if there was a clear guideline, protocol or standard of clinical practice to follow, but much less likely in the context of the children's highly complex conditions and the associated uncertainty:

Res 5:   I think sometimes as nurses and doctors, we perhaps, we're constantly thinking of the answer to what they're asking rather than actually listening to what they're saying.
(Nurse, focus group 4)

Healthcare professionals explained how culture in healthcare that places emphasis on "solution-focussed" or "outcomes-based" measurable approaches affected practice and the acknowledgement of uncertainty:

Res 6:   Well, there is, there's a push to be solution focussed or outcome based and to have concrete outcomes, erm, and erm that's not just, I think it's unfair to say that that's imposed on us, I think that we are people like that, erm, as well. And you know if you look at symptom profiles, the ones that get ignored are the ones that are difficult to manage, like fatigue, loneliness, erm, you know all the kind of things that we can put under "caring", perhaps, you know. And we focus on things like pain management, and, you know drug based pain management, because the non-pharmacological, or the psychological, sociological aspect of it is harder to quantify and manage and provide a quick solution to.
(Doctor, focus group 3)

Furthermore, clinicians were described as having a risk averse, safety conscious approach that could compromise the care they delivered:

Res 8:   I think also with that aspect of measuring risk and safety we actually do risk patient care. Because we are so risk averse and safety conscious that there are whole demographics of patients who suffer, because we can't then do good care.
(Doctor, focus group 3)

There was speculation that some professional groups found managing uncertainty harder than others, depending on their specialty and approach:

Res 13:   So we're very used to medical care, so it's [palliative care], the focus is not just on the medical care, or on the outcome, but the patient experience and the support that that family and child need, the psychological, spiritual, financial, practical needs, so it's seeing the bigger picture
SM: That's quite hard to do though?
Res 13: When you're an intensivist by nature, yeah
(Doctor, focus group 1)

The expectations of professionals about how a treatment or the system should work were also a factor. In the example below, a mother explained that she felt doctors sometimes had unrealistic expectations about how a drug treatment should work, and how they were surprised when this didn't happen, but nurses appeared more realistic:

M011:   And I think doctors are very much optimists. I think doctors are optimists. … It's, "we'll put her on some [drug treatment] and it's going to make everything better", type thing. Whereas I think nurses are realists. I've noticed that nurses will kind of be the ones to say, you know, this might not work and you may have to say… so like a doctor will come to you in the morning and say, "right we're going to look at your blood results and we're going to look at getting you hopefully discharged today". A nurse will come over and say, "we've written out the discharge paperwork, but we're not going to date it because her bloods weren't great today so it might be that"… the nurses are very much realists and the doctors are optimists.
(Interview 3, M011 C011)

Focus group participants described how important it was to be able to acknowledge uncertainty. In the example below, they explained how difficult this could be, but that open acknowledgement of uncertainty could assist with conversations about the possibility of the child dying:

Res 6:   Yeah, I think I don't know whether it's the right thing to do, but when parents ask me what's going to happen in the future, I say I've got a crystal ball sitting on that desk and there's a massive great crack in it. I just don't know. And I use it all the time and I don't know whether it's the right or wrong thing, sorry?

Res 3: *But that concept of uncertainty is the first step towards going to some of those … heavier concepts of actually they really aren't going to survive and then gradually filling in the detail. … You really have to feel your way with each individual family, and it is quite an intuitive process, you have to kind of sense how much they're ready for, reading them, bespoke bits of body language, all the rest of it.*

(Doctors, focus group 2)

Families appreciated discussions about the clinical uncertainty that existed around their child's individual condition and circumstances when they did happen. They expressed frustration when the enormity of their uncertainty had not been adequately recognised or addressed, and preferred healthcare professionals to admit that they didn't always have the answer:

Mother: *If you don't know the [answer to a clinical question], it's probably better to say I don't know, but I'll go and find out. But the barrier of course is time, and the paperwork perhaps, but it shouldn't be that way, you shouldn't give an answer that's wrong. You should say "I don't know". But I don't think it's easy for professionals to say "I don't know".*

(Interview 3, M006)

They described clinicians who would manage their uncertainty with them as "*rare diamonds*"

(M003). These were often more senior members of medical staff:

Mother: *I think the higher up the level, the ladder, the more honest they become in a way. We had a [specialist] who would quite often say "let me think about that, I'll look into it and I'll let you know", when we'd been talking about drug therapies and so on, and [condition] particularly. We've had that. But I think sometimes this thing about, you know, a policy, not to say.*

(Interview 2, M006)

When uncertainty was not openly acknowledged or shared, this could lead to compromised relationships with healthcare professionals. As experts in their child's condition and management, family members were aware of moments where healthcare professionals were trying to provide a more certain solution or answer than was possible. This was which was perceived to be "wrong information":

Mother: *But when you're getting wrong information, and you know it's wrong, and then you've said I don't think that's right, you get looked upon as you're being difficult.*

(Interview 3, M002)

One mother described how her child's distress related to his past life-threatening episodes, and his anxieties related to the unpredictable nature of his condition, were not often part of the conversation with healthcare professionals. As a result, her child (C014) became angry and dismissive of healthcare professionals at times. In this situation, rather than the situation being considered difficult, she perceived that C014 was considered "*a difficult child*":

M014: *The only problem I had in the beginning was getting them to understand that [C014] wasn't a difficult child, that he was just a scared child. So that was my main problem, which I did speak to psychology about and psychology went and spoke to the ward and spoke about his [life-threatening episode]. Why he's so scared of not being able to breathe, I mean the child has been in a mess. So there was a lot of educating of the staff around why he would be crying and wanting me for everything.*

(Interview 1, M014)

Theme 2: The collusion of immortality

Families described an awareness of the severity of their child's condition, but they became involved in a "collusion of immortality" with healthcare professionals, where nobody took responsibility for acknowledging the possibility that the child could die. As described in the previous sections of this chapter, the healthcare system, structured around each pathological system, organ or condition created professional boundaries and fragmentation. Cure-orientated protocols and guidelines were sometimes rigidly followed. Both families and healthcare professionals perceived a lack of holism, and children and families felt their concerns, including the acknowledgement of uncertainty, were not always heard or validated.

Within this system, there was opportunity to avoid open conversations about the possibility that deteriorations in the child's health could lead to dying. Medical management could be directed by a protocol or guideline, or a referral could be made. Focus group participants described the resulting lack of holistic, person-centred care:

Res 14: *They [specialists] find it difficult to look at the child as whole don't they? So certainly in hospital, you very often, you know, they're under cardiology, they're under respiratory, they're under liver, and each one of those can fix the problem. So they can sort the heart problem out, they can sort the liver problem, they can sort the renal problem, whatever, individually, but as a whole you can't, because you can't deal with one without another.*

(Nurse, focus group 4)

In some circumstances, acknowledging the possibility of dying was deferred, particularly at times when the focus of medical management was entirely on life-saving treatments, which were often delivered urgently. The family's awareness of the fragility of their child's condition was particularly prominent during these life-threatening deteriorations. These were not times when acknowledging the parents understanding of how critically unwell their child had become or having any open conversation about the possibility that the child would die was necessarily a priority. The focus was on the immediate and urgent medical management to save their child's life. In the quote below, a mother had realised that her child's condition was life-threatening, stating "*we just thought the worst*". However, the medical management of the acute episode was the priority at that time, and

the opportunity for the parents to express their knowledge that the episode was life-threatening was limited:

Mother: *Then he became really ill. And then I said "this is just not right, perhaps we'll take him in and get him checked". And then the registrar fortunately knew him, didn't she, because she knew it wasn't like him to be lying down flat. And I remember her saying "get him hooked up now, we'll start the [name of drug], the antibiotics". And I remember the nurse saying "why? he hasn't spiked [a temperature]", she [doctor] said "that doesn't mean he won't". And we took him straight up. And he became, within hours, really, really, really unwell. I think we just thought the worst then.*
(Interview 1, M007)

Once the episode had passed, and the child had recovered, life continued and these conversations still did not take place, sometimes because healthcare professionals did not instigate them, and sometimes because families did not wish for them. The culture of the healthcare system was death-denying and death-defying, and this had an impact on the response of both families and healthcare professionals. The possibility of a child dying was a "blind spot", put to the back of the agenda, or coped with through denial:

Res 2: *But I do think there's an element of, when you say that people sort of pretend that people don't die, everyone knows deep down, but there is a lot of denial, or it's put to the back of the agenda, and there is an inability to actually discuss it as a reality. You see that in a lot in adult practice, and you see it a lot in paediatric practice*
(Doctor, focus group 3)

There seemed to be variation in approach amongst individual clinicians:

Res 6: *I think for children under different specialities, you'll get some [healthcare professionals] who are straight in on the, very early on when they'll engage with palliation, and palliative care, whereas others will look for a fix and keep going, and look for a fix. And you're in meetings and you'll hear them, and essentially what they're saying is, you know, you can almost hear the palliative word, but they can't quite do it and they'll keep going, keep going, and it's normally the families then that will kind of ask, "so if that [treatment] doesn't work?"*
(AHP respondent, focus group 4)

The example below illustrates a death-denying scenario from a parental perspective. C009 had been critically unwell and admitted to PICU. This was one of several life-threatening deteriorations. The life-threatening nature of those deteriorations, and the child's underlying diagnosis, had been incredibly difficult for the family to accept. They described the consultant as being "*so negative*", and a need to see *"proof"* of the cause of a previous deterioration, on their terms:

SM: *You know when you said about when [C009] was really poorly in ICU, and they took you into a side room and had that big conversation. Have they ever brought that up again, do they ever talk about it, how poorly he was and..? No.*
Mother: *No.*

SM: *Do you think they should, as a just in case kind of..?*

Mother: *His consultant, she's a bit of a… she's good, but she's always so negative all the time. She always tells you all the negative stuff, she never says anything positive really, does she?*

Father: *No.*

Mother: *She's always really negative about stuff.*

SM: *Right.*

Mother: *And I think if we was to turn round to her and say "well you said he had a [condition seen on x-ray]", she'd still say "well it was", she wouldn't let it rest, would she, even though it wasn't a [condition]. She was like, because she reckons that how really poorly he was at the time, which he was, really, but not as bad as she made it out to be.*

Father: *But she's looking at him from the clinical point of view. Where we're looking for proof more than how he looks. We want to know what's wrong, but they can't say what's wrong because they can't actually see. Because this [x-ray of the condition], there's no image to see the [actual condition], it's just this [sign on the x-ray] or whatever it was. So they're saying it's [condition] because there's [a sign on x-ray] there. But there was no actual, anything there to say yeah, there's a [condition].*

(Interview 3, M009 F009 C009)

Another reason that the possibility of dying was not discussed was the death-defying culture in healthcare. As one mother described, doctors sometimes considered themselves "*masters of the condition*". In this case, it was her experience of the doctors involved in the management of her child's unstable seizure condition. She was aware of how unstable the condition was and the life-threatening potential of the seizures, but there was no conversation about death as a possible consequence of a serious seizure. Instead, the management plan was focussed on gaining control of the seizures through medication:

Mother: *Maybe there's a sort of a "what if" planning conversation could be had then from the point of view of [the condition] to say, this is his [condition], this is how we're managing it. But if the worst was to happen and he would have a big seizure that left him with additional brain damage, what would happen, and at that point you could say, you know, potentially life-threatening, you know, let's have a look at the what if there, that could go there, particular to that condition, perhaps. But they [the speciality team] all tend to be quite… "oh we're going to manage this, we're going to get the seizures down, we're going to" … I don't think they like to have the "what if", they like to think that they're going to get to grips with [the condition] and they like to think that they're going to control the [condition]. And I don't know whether it's the [specialists] that don't want to talk about the "what if", but if they can't control the [condition] … They try to think they are masters of this condition.*

(Interview 2, M006)

Focus group participants described situations where despite the child's condition being highly fragile or life-threatening, clinicians seemed to be provided with opportunity to distance themselves by the way that services were organised, particularly in hospital settings. This could be a change in the rota, for example moving from ward cover to other clinical service work, and no longer having to see children and families on a daily basis, or by making a referral to another clinical team. There was a risk that children and families were left feeling "overlooked":

Res 8: The doctors at the hospital can have a difficult discussion and walk off, and potentially say *"well I don't have to go back there, that didn't go too well, you go back next time". … And actually, it's easier for children to be overlooked I think in hospital, because they can be on the odd week "I'm on hot week next week" or they're "somebody else will take them over" or "that didn't work so let's hand you over to somebody who you like a little bit more", so that's slightly easier when you're in hospital and have all that stuff around you.*

(Doctor, focus group 4)

It was felt that a change of mind-set was required amongst healthcare professionals related to discussing the possibility of the child's death, and palliative care, with families. This change of mind-set sometimes occurred very gradually, and was difficult because it required a change in the nature of the conversations with families from one consultation to the next:

RO11: *If they [consultants] feel it in their mind-set that actually also sometimes helps a consultant think they may be leading to this conversation [related to palliative care] in this consultation, so very different to the last consultation,*

(Nurse, focus group 2)

A difficulty for professionals was that they had their own emotions and feelings of distress related to the possibility that a child, who was their patient, may die. Some appeared unable to come to terms with the fragility of the child's situation and the possibility of the child dying, particularly if there was still opportunity to offer new or innovative medical treatments, which was more socially acceptable and did not require the healthcare professional to acknowledge their own feelings. There was a need for professionals to "have courage" to approach conversations about the possibility of a child dying:

Res 13: *I was going to say however, sometimes there's people who haven't got the courage to do that work*
Res 1: *Sometimes people don't, yeah*
(Nurse and doctor, focus group 2)

Barriers such as "professional boundaries" were described, allowing some professionals to frame conversations about the possibility a child would die as someone else's job, such as a specialist paediatric palliative care team. This also appeared to be a socially acceptable way to manage the problem within the healthcare system:

Res 9: *I think there are huge issues around professional boundaries as well. And I think a lot of people are so concerned about that, that I don't know whether some people maybe, erm, intentionally or non-intentionally hide behind that. That they're the ones, that actually they're the ones not ready to ask those questions or listen to those answers. But the young person is screaming out to be asked those.*
(Nurse, focus group 4)

R003:   *I think the number of people who were referred to the hospice, for respite care, with no mention of, you know, even leave the word hospice out of the discussion if you could, erm and you then you have to introduce that as a concept either in the information that the hospice is setting out or by going to see them with that kind of thing. Um yeah, I think there is an element of that that people feel that "ooh, this might be somebody else's job to bring up", and "I'm not sure that now is the right time or I'm the right person" and there's a certain amount of avoidance.*

(Doctor, focus group 2)

There were concerns about a lack of open acknowledgement of the personal feelings and emotions of healthcare professionals caring for children with life-limiting and life-threatening conditions within the culture of the healthcare system. Healthcare professionals could require professional support (an "ally") in reaching the point where they could firstly have conversations amongst their colleagues, and secondly have these conversations with families:

R011:   *I think it's quite difficult for clinicians as well in that they are often still "fixing". So they need to confront that, you know "I don't want to admit defeat I want to keep going", and actually "I am at this point", and … almost it's we're [the palliative care team are] an intermediary, being that ally as well, in terms of, you know, in that doctors actually being able to have those conversations with each other, erm it's not just with families.*

(Nurse, focus group 2)

Children and families, when describing their relationships with healthcare professionals raised the importance of communication and language. A further potential barrier to discussing palliative care was the word "palliative".

Theme 3: The "p word" problem

As described earlier in this chapter, palliative care was often conceptualised as a distinct and separate service, or phase of a child's care. Children, family members and healthcare professionals in the focus groups, who highlighted the significant impact that this could have on the care and services that children received, described a "p word" problem. The word "palliative" was universally unpopular amongst the children and families, and was a significant barrier to conversations about palliative care or referrals to specialist services. Three families specifically referred to the word "palliative" as a problem, with one mother stating "*don't say the "p word" in front of her [C002], she hates the "p word"*". Sometimes there was a perception that care from the healthcare professionals who had been managing the child's condition would stop if "palliative care" became the management plan:

Res 18: That word "palliative", as soon as someone mentions that word, that's like a door,
       (whooshing sound, indicates door shutting)

(Nurse, focus group 1)

Res 8:   *What we commonly get is, we get, parents who are very upset, because they are told they are palliative, and they feel they are then dismissed from hospital, or dismissed from the specialist, "well, we can't do any more so, hey ho, off you go" kind of thing, erm, and they find that distressing,*
(Doctor, focus group 4)

One family referred to the word "palliative" as "the "P" word" ("*don't say the "p" word in front of [C002], she doesn't' like it*" (Interview 1, M002)). The same mother provided more detail of her experience of the when the word "palliative", and the idea of palliative care, were introduced to her:

Mother: *I'll tell you, even though I'm probably an old thing to it, when we were in the hospital two years ago on her 16th birthday … and they kept on going on about palliative, and the palliative this and palliative that. And there was all these people in this meeting and all I kept on hearing through my head was palliative. So I had to stand up and say, "what do you mean?" And they all just stared at me. And I said, "what do you mean; are you telling me she isn't going to live six months, a year, you need to explain". And they said, "we can't tell you that, we just know that she is palliative". So palliative can mean anything I think from a couple of weeks or days up to a year or more.*
(Interview 1, M002)

"Palliative" had strong associations for others with end of life care, hospices and dying:

Mother: *Palliative means end of life, or life-limiting. Very serious, that's what it means, you're supposed to get that extra support, extra care. That's what it should be, but you don't, that doesn't happen.*
(Interview 2, M003)

Mother: *You'd expect palliative care specialists to be working in a hospice because to me a hospice again is all about that. And I know it's different for children, but it is sort of about end of life. And I know children they talk more about life-limited and life-threatening don't they, life-threatening rather than life-limited, but yeah.*
(Interview 2, M006)

Focus group participants described a highly variable level of understanding of the word "palliative" amongst healthcare professionals, with a need to improve understanding and "*dispel myths*":

Res 17: *I think it's also dispelling the myths with professionals, even the most professional professional doesn't always know what palliative care is, they think its end of life. "Oh I haven't referred because this child's not dying"*
(Nurse, focus group 1)

Res 9:   *People don't like talking about palliative care, but it's because they don't understand what palliative care is. When I got the job and I said to my friends, they were like "oh, like, it's end of life, and just dying people", and I joked with my friends saying "do I need to do a teaching session with you all about what palliative care is?" But they are nurses. They are other nurses here, and at other [city] hospitals, and that's such a barrier to the care that we provide.*
(Nurse, focus group 3)

For some professionals, including allied healthcare professionals, a lack of both undergraduate and postgraduate training opportunities in palliative care was described as a pertinent problem that contributed to a lack of understanding. The idea that palliative care was associated with end of life care pervaded and caused anxiety:

Res 8:   *Actually as an allied health professional, you're not taught it. You don't learn it. You might get exposed if you work in a hospital. And that breeds this whole huge gap between what it is, and if you go to community [professionals] they won't have a clue. The moment you throw a palliative care [request for medication] and then tell them what it's for, they'll have a heart attack.*
(AHP respondent, focus group 3)

Wider societal perceptions and attitudes, including death and dying as taboo subjects, were perceived as other relevant factors, making it difficult to educate and promote palliative care to others:

Res 6:   *From a societal point of view despite everything, the more civilised we become, you know the more invincible we think we are, and so it's you know so death and dying is still really buried. And so it's difficult for those of us who are, who are champions for this to inspire others*
(Doctor, focus group 3)

R013:   *And it's like, it's like the fundraising for our charity, you would think the fundraising for our charity, for a charity that supports children with life-limiting conditions who are going to die, that it would be easy. Actually, it switches people off, not switches people on, people can't go there.*
(Nurse, focus group 1)

Family members of one of the children who was moving to adult services had found that the perception of the word "palliative" amongst staff in adult services contrasted starkly with the care they had been receiving from paediatric palliative care services. "Palliative care" was firmly associated with end of life care and stopping any active care interventions and treatment. It appeared that in the minds of her adult clinicians, continuity her current life-sustaining treatments, as opposed to receiving palliative care, required a commitment to a multiple organ transplant. The child and her family had already considered this in detail:

Mother: *No, no. They are trying to say, the [adult hospital] are saying they don't have people at home on IV [medication] like [C002]. They [people on home IV medication] wouldn't be classed as palliative, one of the staff told me from the [adult hospital] … She come over with a transition nurse, and she was a transition nurse for there. And she said if [C002] didn't want a [multi-organ] transplant, they wouldn't put [C002] on [IV medication] and stuff, and give her IVs. They'd class her as "palliative care" and send her home.*
(Interview 1, M002 C002)

Recognising the "p word" problem, specialist paediatric palliative care team members had given thought to how they introduced themselves to families, as in the example below:

Res 14: *So one of the things, sorry, so when I introduce myself to families I talk about, erm, that I'm a palliative care doctor, partly because if you Google my name, then I will come up as a palliative care doctor, and it's always upsetting if they Google me and then find out. Erm, and I also say that it's a bit different to palliative care for adults which is much more end of life focussed, and I talk about, erm that my job is about supporting families when life is uncertain, rather than just at the end of life, and parents generally get that. And I say that I support them for when life is uncertain, to make the most of life, for however long that is… .*

Res 6: *It is how it's sold, it is how it's sold to a family, if they feel like palliative is because you're giving up, they don't want it. If they think it's part of their journey, so we are still hoping for the best, but planning for the worst, they can take that, they can buy into it, but it is, you see it day in day out, the families that engage are the ones that feel that they're erm they're, it's not giving up at the end.*

*(*Doctor and nurse respondents, focus group 4)

Other specialist paediatric palliative care team members described choosing to introduce themselves using other terms, including the pain management team, or symptom control team:

Res 7: *Sometimes I think the word "palliative nurse" does scare some families, and … sometimes we change the word that we introduce ourselves, sometimes. Pain management, symptom control. Things like that.*

(Nurse, focus group 1)

The provision of "*training and education*" to other healthcare professionals was frequently mentioned as a potential solution, particularly in areas such as communication skills, in order to facilitate the delivery of palliative care more widely. However, there were concerns that only those who were already interested in palliative care would attend. Palliative care was not considered to be an "inspirational" area of practice:

Res 6: *There's nothing inspirational about palliative care, so it's difficult to go out there and sell it to people. You know gene therapy, you'll get loads of bums on seats, if you go out there and say "I'm going to talk about gene therapy". People who are completely disinterested off the street will come along and go "ooh well that's interesting". Erm but if you say "I'm going to talk about death and dying", ok, it'll only be the people who are genuinely interested in it, who are probably already working in it, and then you are preaching to the converted already.*

(Doctor, focus group 3)

A more subtle and nuanced influence on the wider workforce that was occurring alongside formal training, education and integration into other clinical teams, was the role of palliative care teams in leadership and role modelling a palliative care approach to care that included conversations about dying and planning for this, as described in the quote below:

Res 1: *I think an important part of this process is modelling. And if you don't have a service where you are modelling this aspect of care, and actually demonstrating what the clinical benefit is, then you can't grow it. Because your colleagues get such a blind spot … if you don't have somebody doing that type of work, then people aren't going, people don't know about it, or people don't know the experience that it offers.*

(Doctor, focus group 1)

Res 22: *It's supporting the other professionals to be brave enough to sort of look at a child and think "this child needs palliative care".*

(Nurse, focus group 2)

Achieving "buy-in" from other professionals could have a striking impact in achieving "momentum" in terms of developing services, changing attitudes and professional behaviours. This was perceived to be most effective amongst senior clinical professionals:

Res 13: *I'd agree with that, it's about, it's the buy-in of the specialists, of the consultants, it's at that level, if you've got that buy-in at an early level, and it's right in at the start of that journey, as you say, it's just considered part of the normal practice then. It's when it's brought in as coming in at the end, and then it's seen as palliative, and it's seen as big, a really big deal, as opposed to this is part of your rest, your respite, this is part of your support, and you may be discharged.*

(Doctor, focus group 4)

Focus group participants expressed feelings that their ambitions to improve palliative care for children was poorly understood and unheard by service managers and commissioners, often because there was no extra funding provided. Achieving the support of senior management was a key factor in securing funding for the development of services:

Res 2: *I think the other thing is that was a really big factor, is that we had a Chief Exec, who was really really supportive of palliative care. And virtually anything we asked for they would find a way to get it for us*

(Doctor, focus group 3)

8.4. Summary of chapter

A concurrent process of learning to navigate the healthcare system, processes and procedures accompanied the family's continual adaptation and learning about the management of the child's condition. Accessing and co-ordinating healthcare services within a fragmented, specialist system, caused families to feel that "*everything's a fight*". Feeling that there was a fight for their child's needs provoked a range of (often antagonistic) amongst family members, but not necessarily the children, who tended to live in the present. A biomedical approach that focused on medical solutions or "fixes" to problems added to the complexity of the child and family situation. The short and long-term uncertainty that the children and families lived was not always acknowledged, and a

"collusion of immortality" was created in a healthcare culture that was death-denying and death-defying. The fragmented biomedical system, focussed on cure, provided opportunity to avoid conversations about the possibility of death. Underlying all of this was the thought that a child may die being unbearable.

Palliative care tended to be conceptualised as a separate and distinct service, the introduction of which could be difficult at least in part due to the term "palliative". Children and families reported a range of different care needs being met by palliative care teams. Wider acceptance of palliative care as an entity, approach and specialist service, across the healthcare system appeared necessary. Specialist paediatric palliative care professionals, as members of a relatively new specialty, expressed frustration at the inconsistency in services and training opportunities, and perceived multiple barriers in terms of future development of the speciality, particularly related to financial resource. Policy makers and system leaders who were committed to improving in palliative care were key to developing services.

9. Findings 3: Application of a realist logic to the findings

9.1. Overview of Chapter 9

Chapters 7 and 8 have presented the findings of the research at different system levels. The findings described the children's experiences of living with a life-limiting or life-threatening condition, the impact this had on their family members and the implications for family life. At the micro-system level, their interpersonal relationships with individual healthcare professionals played a key part in their experiences of healthcare. Findings at the meso-system level revealed challenging relationships with the healthcare system, a situation recognised by the healthcare professionals who took part in the focus groups. Macro-system influences, including culture within healthcare organisations and mixed views of the term "palliative" further complicated these relationships.

In this chapter, a realist logic has been applied to the findings of the thematic analysis. The content of the themes has been scrutinised to propose which themes, subthemes or content function as contexts and which as outcomes. The mechanisms that connect these contexts and outcomes have then been abstracted to devise context-mechanism-outcome configurations (CMOCs), which provide explanatory descriptions of how outcomes are produced through the activation of hidden mechanisms. As a reminder, table 9.1 provides the definitions of contexts, mechanisms and outcomes:

Table 9.1: Glossary of realist terms (Adapted from Papoutsi et al (87))

Term	Explanation
Context	Pre-existing structures, settings, environments, circumstances or conditions that influence whether or not certain behavioural and emotional responses (i.e. mechanisms) are triggered.
Context-Mechanism-Outcome Configurations (CMOCs)	Describe the causal relationships between contexts, mechanisms and outcomes i.e. how certain outcomes are achieved through mechanisms being triggered in certain contexts.
Mechanisms	The behavioural or emotional response which is triggered in certain contexts. Mechanisms are context sensitive and are usually hidden.
Outcome	The impact of mechanisms being triggered in certain contexts.
Programme theory	A set of theoretical explanations about how a particular programme, process or interventions is expected to work.
Mid-range theory	Theoretical explanations which are suitable for testing through further research. A programme theory can be specified at the mid-range.

The chapter is presented in four sections. The first section applies to the child and family situation, and the following sections describe the overall "programme" of palliative care at micro, meso and macro-system levels. The first section provides CMOCs drawn from the empirical research relating to the micro-system. The second section provides CMOCs that relate to the child and family

interactions with the healthcare system, their experiences of palliative care (meso-system), and the influence of wider culture (macro-system).

The findings provide evidence to address the following research questions:

- What outcomes are important to children and their families?
- What are the mechanisms by which these desired outcomes are achieved?
- What are the contexts that determine whether or not these mechanisms produce the intended outcomes?

9.2. Contexts and outcomes in the child and family situation

The empirical research has provided rich, in-depth insights into the child and family situation. The data is important because it describes the child and family situations that become the contexts in which palliative care must be delivered.

Micro-system CMOC 1: Family adaptation

Key moments for the family were the child developing a condition which is life-limiting or life-threatening and from which they may die (context), and the process of realisation for the families (mechanism). For every family, there was a continual process of adjustment (mechanism). The precariousness of the child's condition, over which they had no control, with unpredictable, sudden deteriorations requiring urgent intensive medical treatments (contexts), triggered further moments of adjustment and adaptation (mechanisms). With every change in their child's condition (context), family members framed and re-framed their hopes and expectations (outcome). Due to the nature of the child's condition, both the children and family members had implicit knowledge that the child could die (mechanism), but this tended to be held in their minds (outcome), and alluded to in conversations rather than openly discussed.

A strong theme related to family adaptation was that the situation was not one that had come about through choice (context). A child having a life-limiting or life-threatening condition was a significant imposition on family life; they were "cornered" by the arrival of the condition. The constant process of adjustment and adaptation was one that the children and their families were obliged to face (mechanism). They could not maintain control over what was happening and were therefore in a position of vulnerability, disempowered (mechanism) by the impact of their child's condition both on the child and on their family life.

Healthcare professionals provided advice and written resources; however, much of family member's learning happened through observation and experience (mechanism). They also sought support from other families in similar situations, and, in turn, provided advice and support to others. Over

time, they became experts in their child's condition, medical treatments and the impact of the child's condition on the family (outcome). The process of adaptation and learning, gaining expertise and continually re-framing their hopes and expectations, along with the fact that this was not a choice, all required dedication from family members and resulted in the child's condition becoming a family vocation (outcome). The contexts, mechanisms and outcomes described in CMOC 1, related to family adaptation to their situation are represented in Figure 9.2.1:

Figure 9.2.1: CMOC 1 - Family adaptation
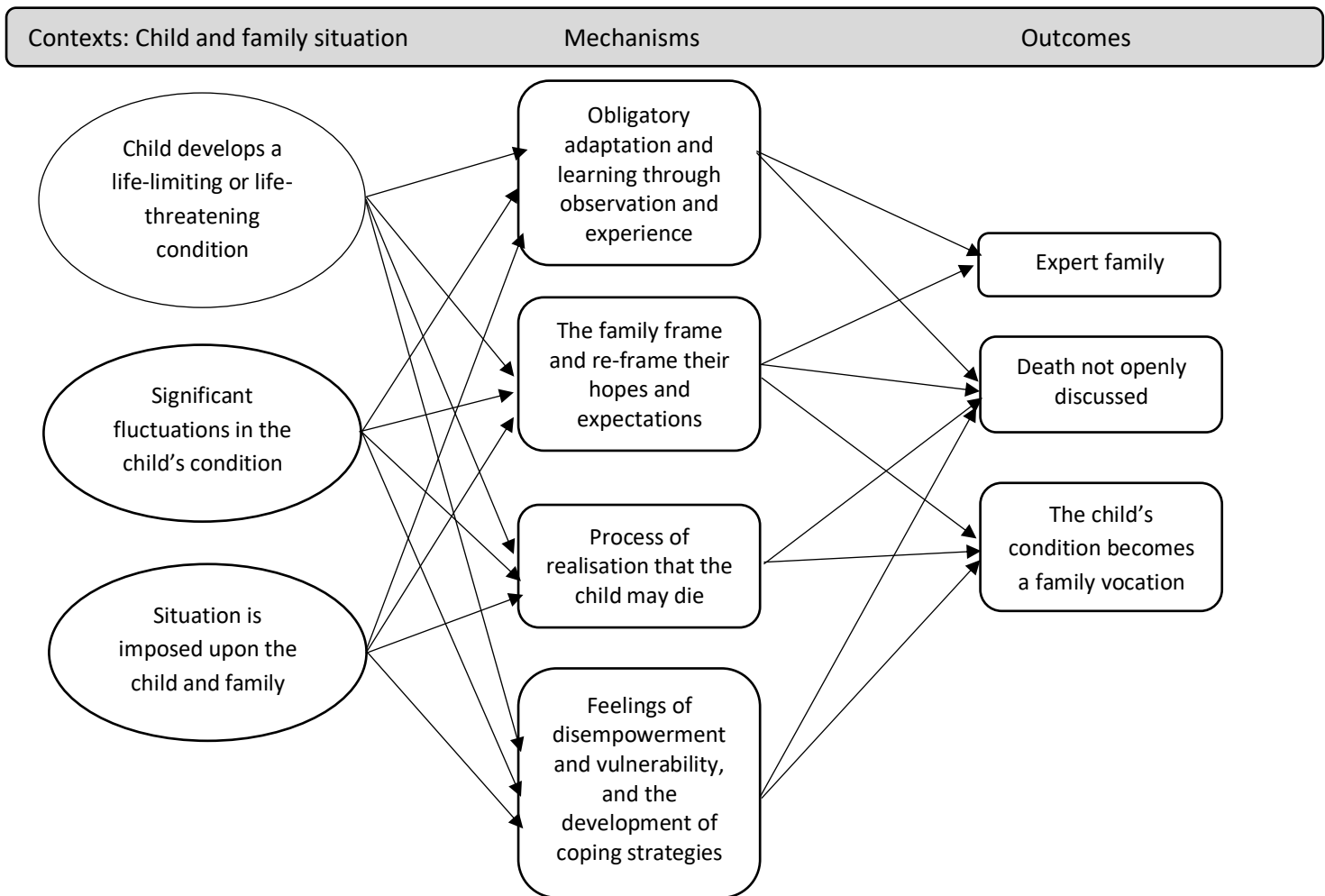
Micro-system CMOC 2: The child's condition as a family vocation
A life-limiting or life-threatening condition in a child, and the significant fluctuations in the child's condition that followed, inflicted a significant change in the parental role (context). The role of parent carer extended beyond the parental responsibilities usually associated with raising a child, to attending to their daily healthcare and nursing needs. For some, these were highly complex, with the use of life-sustaining medical technology, such as long-term ventilation and enteral feeding

equipment at home (context). Treatments and interventions, such as the administration of intravenous fluids and medication were also delivered at home. Healthcare professionals taught parents the necessary skills and competencies (context). The continuous process of learning through observation and experience continued (mechanism), and family members developed expert knowledge of both the healthcare and personal needs of their child (outcome). The child's condition, associated symptoms and treatments, caused anxieties and concerns which family members, particularly parents, had to cope with (mechanisms). They had dual responsibilities: to manage the practicalities of their child's care needs, as well as the emotional impact. This became part of everyday life and a "new normal" (outcome). The change in parental role affected the parents' sense of self and personal identity (mechanism), and they had to draw on their personal resilience and coping strategies (mechanism) in order to take on these dual roles.

There was a desire amongst the family members who took part in the research to "get on with it", even at the most difficult times (mechanism). Their homes became healthcare systems (outcome), which parents managed (outcome), constantly negotiating and re-negotiating with other systems to ensure that the needs of the family were met (mechanism). This included basic needs such as grocery shopping, and disposing of household rubbish. At home, in this system, parents were in charge of making things work. This required dedication, high levels of organisation and often assertiveness, which were all part of their expertise (outcome).

The lack of choice for family members was a pervasive theme. The child's condition became a vocation, but not through choice. At times, this vocation was all consuming. Furthermore, at moments when their child's condition was life-threatening, they had no choice but to hand over care of their child to healthcare professionals, which required a great deal of trust. This was a position of further vulnerability and disempowerment, and was at odds with the requirement for parents to take control at home. Family members required resilience, but given the context of their child's condition and the associated disempowerment and vulnerability, this resilience was fragile (mechanism). These contexts, mechanisms and outcomes, describing the child's condition as a family vocation are outlined in Figure 9.2.2.

Figure 9.2.2: CMOC 2 - the condition as a family vocation

Contexts: Child and family situation	Mechanisms	Outcomes
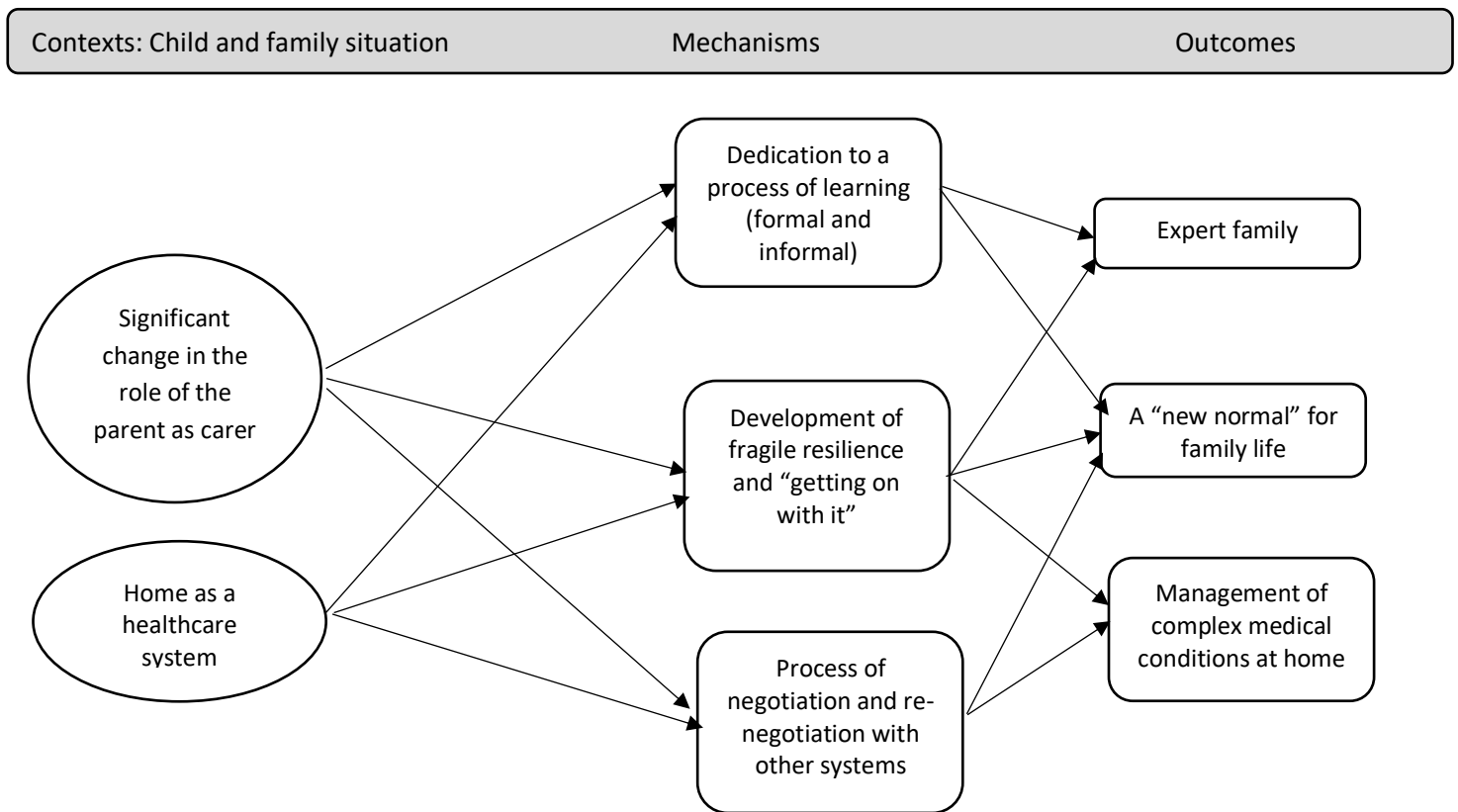
Micro-system CMOC 3: The impact of the condition on the child's life

The children who took part in the study all had an awareness of their condition, including acute and insidious fluctuations, regardless of their age (context). Their lives were dominated by their condition and treatment plans (context). Their parents tended to become their trusted spokespeople and surrogate decision makers (context). They displayed an acceptance of their condition as part of life (context). Most were aware of the potentially life-threatening nature of their condition, and had spoken about this in some way with their parents.

The children all had their own interests and priorities and it was very clear during the interviews that they did not wish to be defined by their condition (context). They were passive recipients of the decisions that were made by the adults around them in terms of both their medical care and their social activities (mechanism). Given the precariousness of their situation, and that their treatments and medical management plans could dominate their lives, there was a risk that their own priorities were not heard or addressed (outcome). When the children felt that their priorities were unheard, or that they were being in some way defined by their condition, this had an adverse impact on their psychological wellbeing (outcome). For example, C007 expressed pain at a time when he was receiving intensive cure-orientated treatment. His parents recognised the large psychological element of the pain, but felt this was not addressed adequately. At times, the children expressed

anger and frustration. C007 described a difficult experience in the emergency department, directing his frustration at staff and stating, "they don't know how to care". Some of the children became ambivalent about decisions regarding not only their healthcare but also other activities that might be considered by others to be unique and privileged experiences, such as C008, who expressed ambivalence towards her trip to the Houses of Parliament (outcome).

The children were keen to see friends and had a desire to maintain normality (mechanism) by joining in with activities such as going to school, the cinema and cafes. C014 wanted to go to the "All you can eat" buffet, despite his enteral feeding, and C009, who was bed bound at the time of the interview, wanted to be able to go outside and play, because he "loved that". Sometimes these needs were unheard or heard but not considered practical, due to the impact of the child's condition. Being unable to join in with activities that they enjoyed had an impact on their psychological health and wellbeing (outcome).

For some families, the child's interests and priorities were considered a pressing issue because there was a feeling that time with their child may be limited (mechanism). This was often informed by their previous experiences of their child's life-threatening deteriorations, when the possibility of death was acute. There was a desire to live life to the full, making the most of every opportunity, while this was still possible (outcome). The situation was exemplified by one child who had been "horse riding in Jimmy Choos" (C002), seizing the opportunity to do so because she was alive after many life-threatening deteriorations and admissions to intensive care. The contexts, mechanisms and outcomes described in CMOC 2 are outlined in figure 9.2.3:

Figure 9.2.3: CMOC 3 – The impact of the condition on the child's life
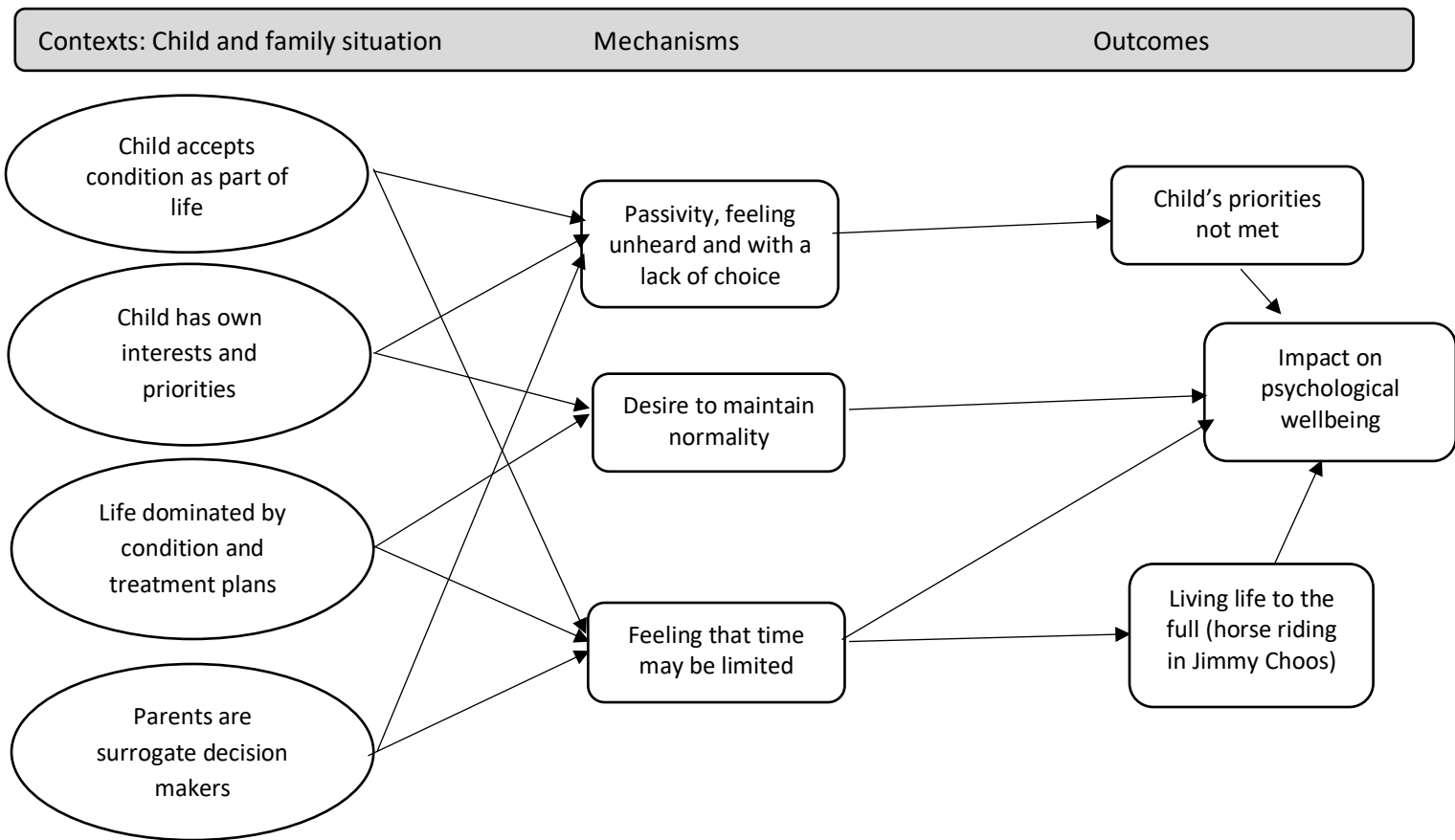
9.3. The micro-system

Micro-system CMOC 4: The child's relationships with healthcare professionals

The children's relationships with healthcare professionals were highly variable, and depended on where the children were and which professionals were present at the time (contexts). On the ward, it was nursing staff, junior medical staff and housekeeping staff who were spoken about by the children, since they were seen and interacted with more frequently (context). Senior medical staff who were seen during outpatient appointments, and other healthcare professionals such as community nurses, were more prominent in the children's minds when they were living at home (context). Healthcare professionals became associated with different care environments (mechanism), sometimes with an expectation that they would be there (mechanism). It was important to the children that healthcare professionals were accessible and responded in a timely fashion (mechanisms) to their needs or requests, such as the need for pain relief.

The children wanted to feel that they were being "looked after" and secure (outcome). This involved feeling valued as individuals, and familiar healthcare professionals providing their care consistently (context), particularly those who knew the intricacies of their condition and treatments. It was also important to the children that the needs of their families were being met (outcomes). The

mechanisms that underpinned these relationships included a kind and compassionate approach on behalf of individuals, and the development of trust (mechanisms). Professionals who "brought tea" and "made us laugh" were valued. Relationships with some healthcare professionals were viewed as friendships, particularly those who shared personal interests with the children, who were consistent and reliable, and who spent time with their family members. These relationships were achieved through consistency (context). The children described individual healthcare professionals positively when they knew them and trusted them to carry out invasive procedures, such as cannulation or phlebotomy. Problems arose when this didn't happen. C008, for example, would object to being seen in clinic by a phlebotomist that she didn't know, who might use a different needle for the procedure. C009 and C011 were more forgiving, expressing an appreciation that healthcare professionals were doing the best they could. Figure 9.3.1 outlines the contexts, mechanisms and outcomes described in this CMOC:

Figure 9.3.1: CMOC 4 – The child's relationships with healthcare professionals
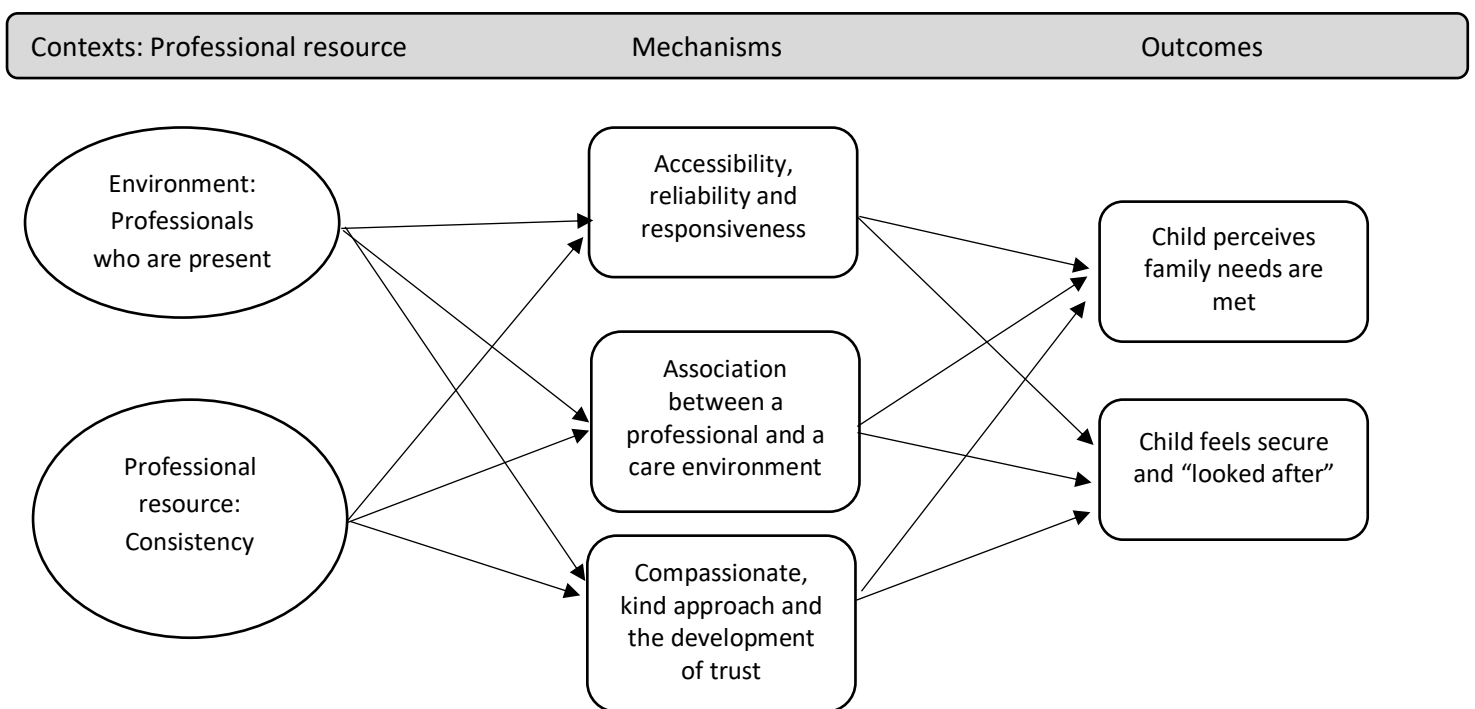
Micro-system CMOC 5: The family develop key interpersonal relationships with healthcare professionals

The interpersonal relationships that developed between individual healthcare professionals and family members were critical. Two key contexts were prominent in the findings of the interviews and focus groups both of which related to professional resource: consistency of healthcare professional within an environment or a team, and a motivation within individual healthcare professionals to provide a holistic approach to care.

Consistency (context), achieved through relational continuity or through repeated encounters in a particular environment, allowed professionals to gain insights and knowledge of the individual child and family situation (mechanism), with all of the associated complexity and uncertainty. This knowledge, together with a perception that the healthcare professional had respect for the family situation, led to trusted, authentic relationships (outcome). Moments when healthcare professionals advocated for the child and family within the healthcare system added to the development of trust and on occasions could be life saving for the child, for example, when the child was in PICU and a healthcare professional who was aware of the child's life at home could contribute their knowledge to medical decision making.

The motivation of an individual healthcare professional to deliver care in a holistic fashion, with consideration of palliative care need, was a significant factor in the care that families received (context). It is described here as an important context because the data from the family interviews provide evidence that this was not a universal approach. Where this professional context was present, children and families perceived that professionals had a kind and compassionate approach to care (mechanism). They experienced support (outcome) through healthcare professionals being willing to "be alongside" them at the most difficult times (mechanism). Sometimes this was just sitting with family members, without speaking or trying to find a solution to the situation. Several families recognised how supportive this action could be (outcome), as did the healthcare professionals who took part in the focus groups. Quotes from the family interviews highlighted the value of professionals "being alongside" families at difficult times (mechanism). Often no conversation or suggestions from the healthcare professional were necessary; there was a feeling of support for family members generated by the healthcare professionals being there. If there were difficult moments when no one was available to be alongside family members, this could heighten the level of distress that they experienced.

Other compassionate acts included healthcare professionals noticing and commenting on details that referred to the individuality and personality of the child. These actions and ways of providing care underpinned outcomes including the families feeling listened to and heard (outcome), and a feeling that they had healthcare professionals around them who cared and who could share their emotional burden (outcomes). Families would describe these individuals as having commitment "over and above" their usual professional role.

The motivation of individual healthcare professionals to deliver a holistic approach to care to children and families triggered another underlying mechanism, an innate ability to bear witness to the child and family situation, with an ability to acknowledge that the death of the child was possible

(mechanism). There was "mutual investment" in the relationship between the child, family and healthcare professional, related to their shared experiences and the trust that accumulated between them over time (outcome). Healthcare professionals who took part in the focus groups recognised this situation, but also described the level of vulnerability (mechanism) that they experienced in the development of these relationships, which could affect both their professional and personal lives (outcome). These contexts, mechanisms and outcomes are outlined in figure 9.3.2:

Figure 9.3.2: CMOC 5 - Key interpersonal relationships with healthcare professionals
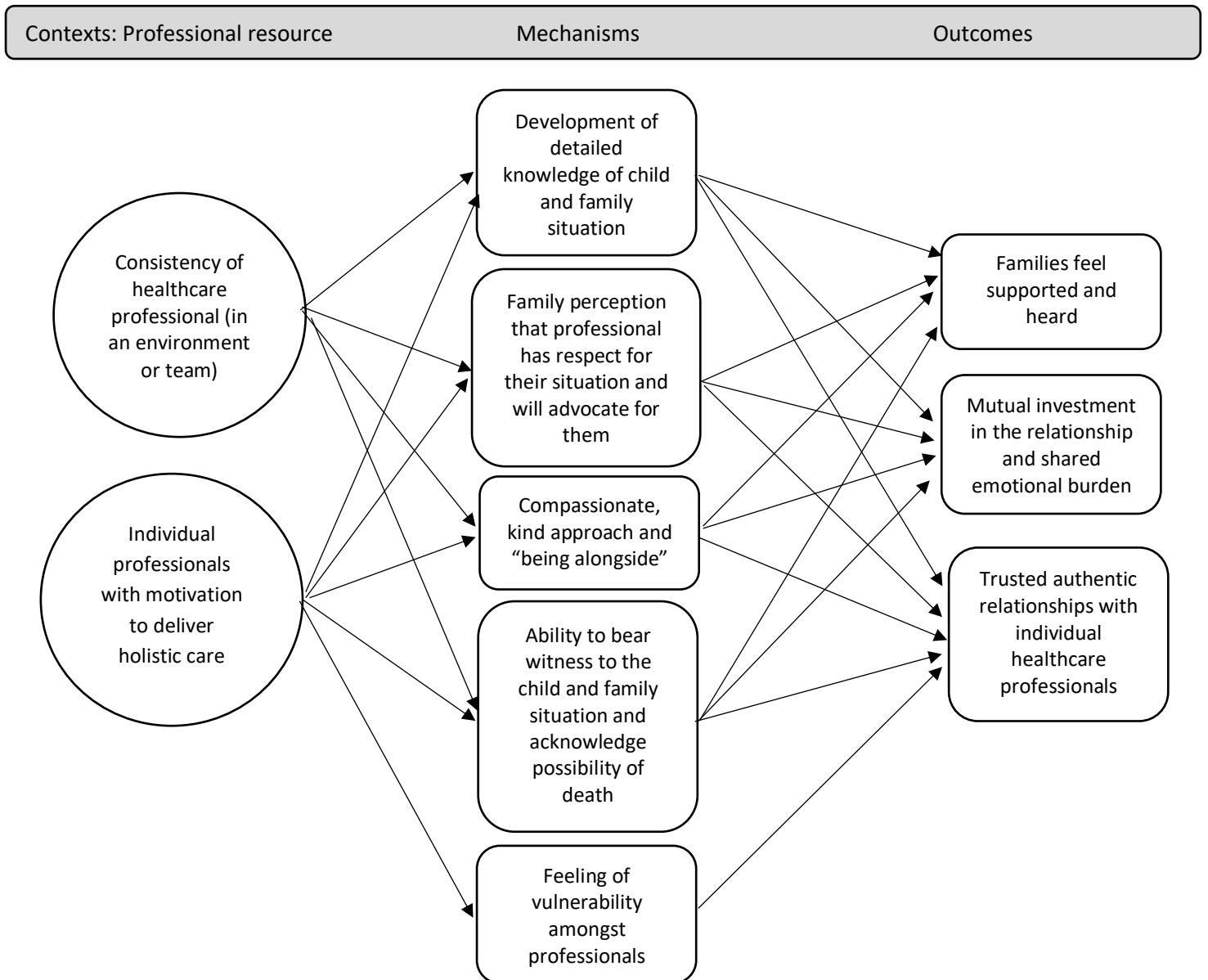
Micro-system CMOC 6: Death is almost impossible to acknowledge

The families lived with the possibility of their child's death as a constant presence. As soon as it became apparent that their child had a life-limiting or life-threatening condition, the possibility of death appeared (context). Death and dying were rarely spoken about during the interviews; they were alluded to in conversation rather than being discussed openly. During periods where the child's condition was stable, the possibility of dying and conversations about death were side-lined, while the child and family got on with managing day-to-day life. When the child became acutely unwell, or when they were receiving disease-orientated treatments, the focus was on managing this, rather than on what would happen if the child's condition deteriorated.

The fragility of the child's life and the possibility of death became apparent during each fluctuation, serious deterioration or relapse in the child's condition (context). At these times, parents had no choice but to hand over the child's medical management to healthcare professionals. They would follow the lead and instructions of healthcare professionals, and admitted that they did not necessarily realise the severity of the child's condition until afterwards. M001, for example, recalled healthcare professionals recommending an admission to intensive care for C001 as a "need for a bit of extra monitoring". It was only retrospectively that she had realised the implications of the admission, and how critically unwell C001 had become. C001 had subsequently told her that he had thought he was going to die during that admission to intensive care. Other parents admitted to thinking "the worst" (M007) at these times. They described feeling "petrified" (M002) and "terrified" (M011), and drew upon their personal (but fragile) resilience, employing sophisticated coping strategies including denial and needing to think positively (mechanisms).

The death of a child appeared to be a distant concept, impossible to acknowledge (outcome), even for those families who had made extensive Advance Care Plans (ACPs). The families views were influenced strongly by their previous experiences, observation and learning. Life-threatening deteriorations, with intensive care admissions, had become a normal part of life for many of the children and families, and a period of recovery following these deteriorations was expected. This was at least in part due to the approach taken by healthcare professionals at those times; the situation was familiar and normal to healthcare professionals as well as to the children and families. Death became a distant possibility because the children repeatedly made a recovery. This pattern of deterioration and recovery lent itself to a situation of mutual pretence (outcome) between families and healthcare professionals, with death existing as an abstract and distant concept, and the severity of the child's health deteriorations becoming part of normal life.

The families had highly variable levels of acceptance of the possibility that their child could die. One mother (M003), whose child had had an ACP for many years stated that as a family, they would "*never think of taking the ACP*" with her when her child was admitted to hospital with a serious deterioration in her health. This was partly because serious deteriorations were a common occurrence, so part of normal life for the child and family, but also demonstrated how important the family coping strategies, including denial, were when the possibility that their child would die was a constant presence in their lives (mechanisms). The fact that the family had developed an ACP demonstrated some acceptance that their child would die eventually, and a willingness to comply with paperwork and processes, but this was not a possibility they could contemplate on an everyday basis (outcome).

Some parents were actively in denial about the possibility of death. One set of parents described their conversations with a consultant about the possibility of their child dying as "*she's always so negative*" (M009), dismissing the possibility so that they could continue to cope and manage the impact of their child's condition and care on their family life. The thought that the child might die was abhorrent and unbearable (mechanism). In this scenario, concerns about the possibility that the child would die raised by their healthcare professionals were too painful, impossible to face, and were therefore dismissed (outcome).

All of the children except one displayed unwavering trust in the adult-led care and treatment decisions made on their behalf. As described in CMOC 3 above, they were passive recipients of the decisions made by the adults around them (this is an important context). They trusted that decisions would be made to keep them alive and that the adults around them would care for them, come what may. They displayed some pragmatism about the possibility of dying (mechanism), with one child, aged 17 at the time of interview, openly discussing her ACP and funeral plans (C002). She was the only child who took part in the study to do so. She was a young person who had survived multiple significant life-threatening deteriorations, and whose family and healthcare professionals had supported her to take part in ACP discussions (outcome). She had experienced bereavements of her own, and displayed some acceptance of death as an inevitable part of life (mechanism). The adults around her shared the emotional burden associated with contemplation of dying (mechanism), which made proactive care planning possible.

One of the children had significant anxiety related to the degenerative nature of his condition and the possibility of dying (outcome). His mother, who was well informed about his condition, accepted that there was an inevitable deterioration towards death, was aware of the level of distress that he was experiencing related to the possibility of death, and was proactively seeking psychological

support for him (outcome), although there was a perception that the level of support available was inadequate. Another child (C009), who was undergoing intensive cure-orientated treatments, had tried to articulate his feelings or need to talk about death by telling his parents that he wanted to die. This had caused his parents, who had dismissed dying as a possibility in order to cope, to experience further distress and refer to his thoughts as "silly". There was clear conflict between the perspective of C009 and his parents, who he generally tried to please and protect (mechanism). Psychological support had been sought (outcome) for C009 at this time, but again had not been perceived to have provided much benefit by his parents.

These complex CMOCs are outlined in the two diagrams below. Figure 9.3.3 outlines contexts, mechanisms and outcomes that can lead to situations where death is impossible to acknowledge. Figure 9.3.4 demonstrates contexts, mechanisms and outcomes that are present when some acceptance of death as a possibility has occurred, particularly from the perspective of the children:

Figure 9.3.3: CMOC 6.1 - Death is impossible to acknowledge
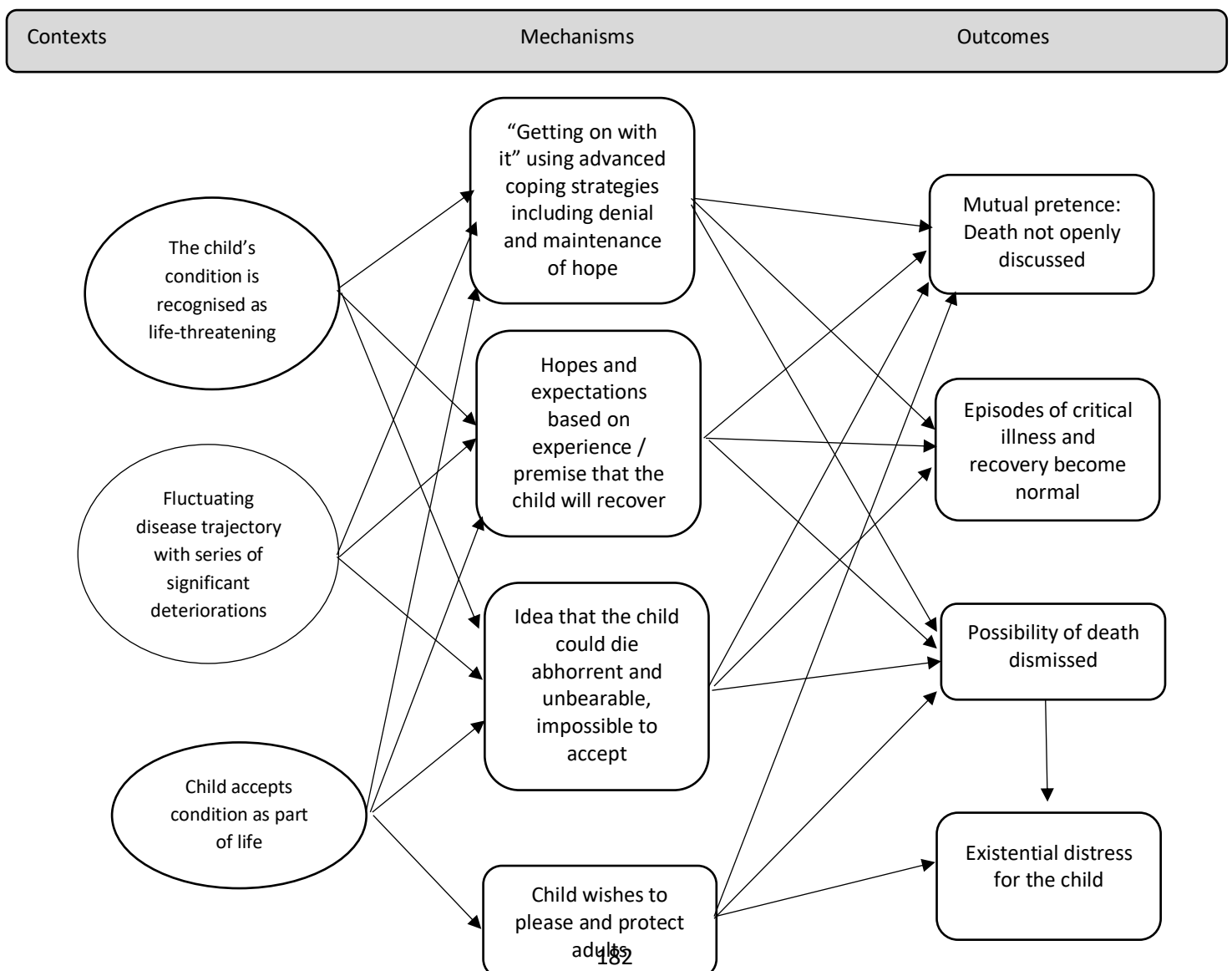
Figure 9.3.4: CMOC 6.2 - Death is acknowledged as part of life

| Contexts | Mechanisms | Outcomes |

Child accepts condition as part of life

Child is passive recipient of decisions made by adults around them

Pragmatic approach to dying

Acceptance that death is inevitable part of life

Unwavering trust in adults

Adults share emotional burden

Ability to take part in end of life care planning

Impact on psychological health
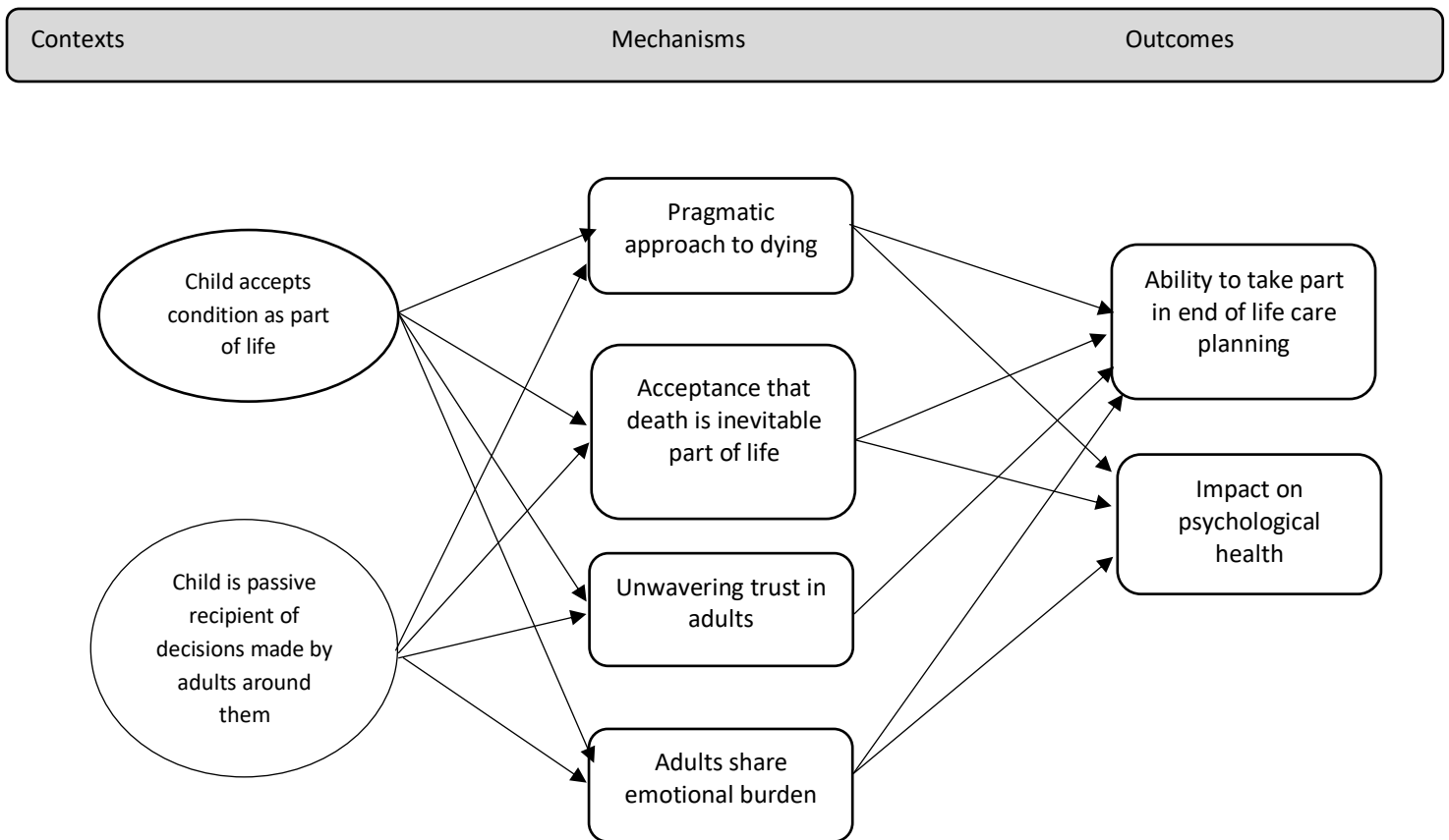
9.4. The meso and macro-systems

The child and family relationships with the healthcare system were complicated, partly due to the complexity of the child's condition, which required the input of several specialist teams, and partly because of the organisation of the healthcare system. Families experienced a healthcare system that was fragmented and rigid, and could be a cause of confusion and anxiety.

Meso-system CMOC7: Relationships with the healthcare system
Regardless of the condition, the children's healthcare tended to be managed by several different paediatric specialists and their teams, who were mainly based in one of several acute hospitals (context). For some children, this was because they lived with a range of long-term conditions that affected different body systems or organs. For others, the side effects of treatments or the effects of their condition resulted in new problems that required the addition of a new drug treatment or referral to another specialist team (context).

Although specialist management was highly valued, there were frustrations related to achieving continuity of care (context). There was compromised continuity within each specialty due to rota or service changes, when clinicians who had been caring for the child and their family for some time

moved on to another job or element of service delivery. Families experienced uncertainty and anxiety related to these changes (mechanism), and described feelings of sadness, frustration and abandonment (mechanism).

When several specialist teams were involved in the child's care, communication between each of the different teams was often poor, particularly when the teams were located in different healthcare settings. Families did not assume that important care decisions made by one team would be communicated to other teams. They expected delays in communication, for example if the treatment plan was written in a letter. M006 provided an example of having to ensure that the specialists who took care of her child's upper limbs sent copies of letters and treatment plans to those involved in the care of his lower limbs, even though they were based in the same hospital. Communication with primary care about changes in prescriptions were particularly problematic. The practicalities of managing care delivered within this fragmented system where communication was poor, was extremely challenging (context). Receiving care from several different specialist teams required a high level of organisation, and parents would often take on the role of care co-ordinator (outcome), drawing on their coping strategies to manage while at the same time experiencing feelings of anxiety and frustration (mechanisms).

Both family members and healthcare professionals described the impact of an evidence-based guideline and protocol driven, risk-averse culture in healthcare on the care that the children received (context). They had experienced approaches to care which resulted from the rigid application of guidelines and protocols, with the ideas, concerns and priorities of the child and family left unheard and unacknowledged (mechanism). Often these concerns were based on the family's expert knowledge of the child's condition (context). Families described the approach, which prioritised guidelines and protocols over their knowledge of their child's circumstances, as lacking "common sense". It would cause them to feel that the needs of their child as a person rather than a condition, were unrecognised, which had a negative impact on their perception of care (outcome). Advocating for their child's needs and priorities in this system was a further motivation to take on the role of care co-ordinator (mechanism).

Referrals to different specialist teams for new problems on the basis that there was a medical explanation for each problem assumed that some further medical treatment was possible (context). Healthcare professionals described this approach as "listening to fix" (mechanism). "Listening to fix" could cause families to feel unheard, particularly if they experienced a series of "hand-offs" between teams, with no single specialist or team taking responsibility for management of the uncertainty (outcome). Focus group participants referred to this as the existence of "professional boundaries";

the specialist system allowed professionals to, consciously or unconsciously, create boundaries, on the basis that a particular problem was outside of their area of expertise (mechanism). While this may have been the case, there were many medical problems that the children lived with that did not have a clear medical explanation and were associated with clinical uncertainty, so did not have a clear solution regardless of the number of specialists involved. Shared and informed decision making became a major challenge where significant clinical uncertainty clashed with the biomedical model of care and professional norms that involved "listening to fix", applying protocols and guidelines, and continually developing new medical treatment plans.

As outlined in the micro-system CMOCs, healthcare professionals who listened, took into account the child and family situation and their views, demonstrated respect for the family expertise and shared the emotional burden, stood out for families. When this occurred, the child and family felt supported by trusted healthcare professionals and that their burdens were shared. At a meso and macro-system level, organisational influences and culture constrained healthcare professionals and their ability to interact with families in this way (mechanism). Regular system changes which resulted in a lack of continuity of care, coupled with a biomedical culture where "listening to fix" was the norm, with onward referrals and more medical treatments being regarded as the solution, came at a cost to the psychological and supportive elements of care, the need for which was keenly felt by families.

These contexts, mechanisms and outcomes are outlined in figure 9.4.1. The diagram compares child and family contexts and mechanisms, with relevant healthcare system contexts and mechanisms. The outcomes included in the diagram all relate to the child and family experience.

Figure 9.4.1: Relationships with the healthcare system
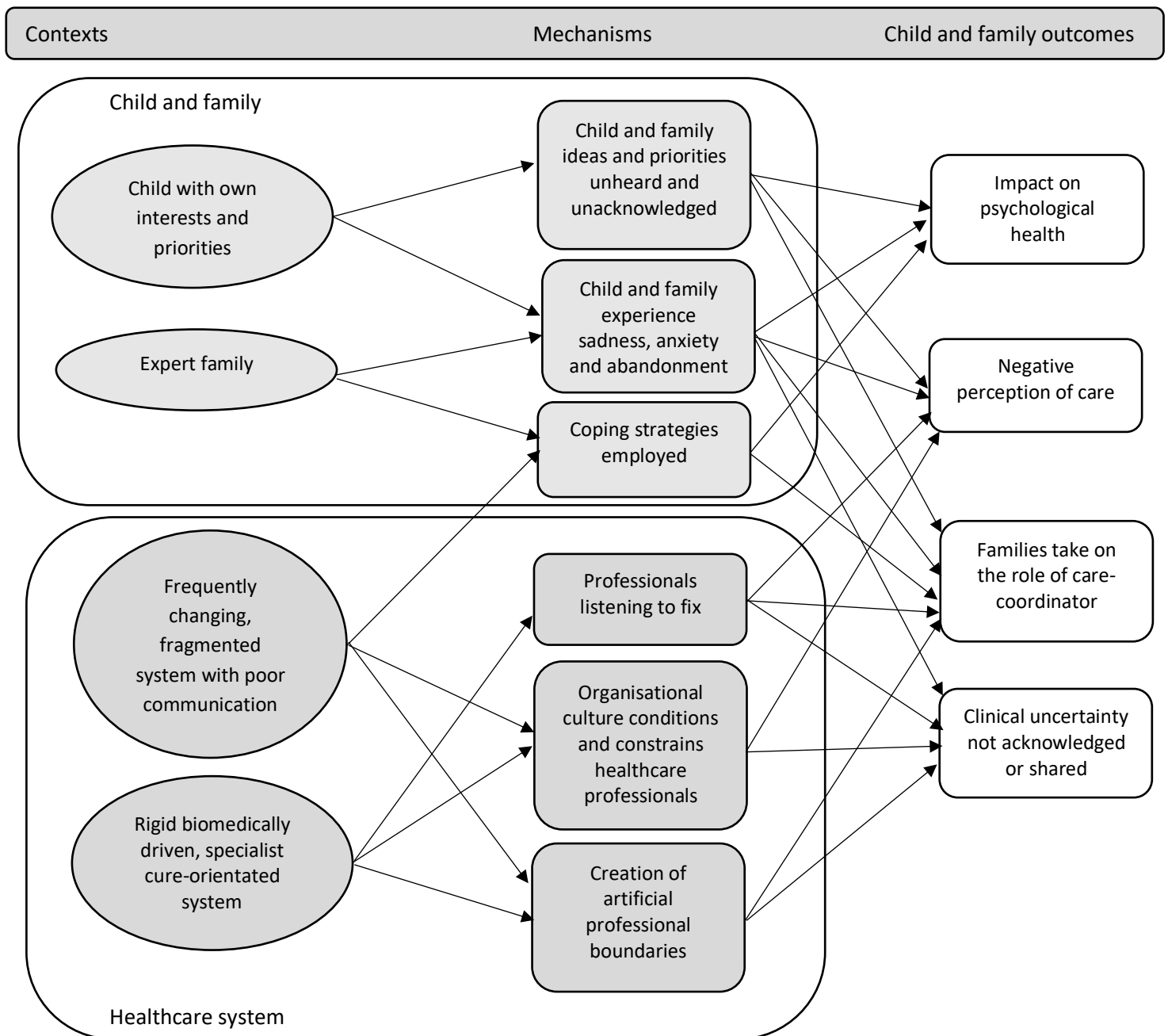
CMOC 8: The collusion of immortality

CMOC 6 described a situation where children and families lived with an underlying, often unspoken knowledge that the child could die from their condition. CMOC 8 outlines the influence of the healthcare system on that unspoken knowledge. It describes how the children and their families became involved in a "collusion of immortality" (outcome).

The biomedically driven healthcare system in which the children and families received their care (context) provided multiple opportunities for intrusive technical interventions to support life, but

tended to avoid open conversations with the child and family about death. Furthermore, the possibility of death in a child was a devastating prospect, against social and cultural norms, both within healthcare organisations and wider society (context).

Difficulties acknowledging clinical uncertainty, underpinned by a biomedical culture that placed emphasis on evidence-based guideline and protocol driven care, led to an expectation amongst professionals that the outcomes of treatments or the disease could perhaps be predicted. M011 described doctors as "optimists", who "can't believe it" when a medical treatment doesn't work as they had expected it to or hoped it would. This clinical culture, with a clear focus on cure, sought certainty (mechanism). Clinical uncertainty was difficult to face, and often unacknowledged. Protocols and guidelines provided a framework for clinicians to refer to in order to maintain some certainty (mechanism). The possibility of discussing a situation where a child might die was extremely difficult (outcome). Focus group participants referred to this, pointing out that many healthcare professionals with a career in paediatrics expected to "make children better". The idea that a child's condition is incurable, or that the child could die of their condition, may be an unbearable thought for some (mechanism).

This situation could lead professionals to avoid having discussions with children and families about the possibility of death by making use of strategies such as onward referrals to other medical teams, or by rigidly following protocols and guidelines (outcome). Onward referrals to other specialist teams allowed for conversations about dying to be "handed-off" from one team to another. In the (unacknowledged) situation that an individual healthcare professional could not bear the thought that the child could die (mechanism), it was perhaps more acceptable to define a professional boundary (mechanism), placing conversations about dying beyond the expertise or remit of an individual or a specialist team, deferring these and hoping that another professional or team would take responsibility.

The conceptualisation of palliative care amongst healthcare professionals as a separate entity; either as a distinct phase in a child's condition, or as a specialist service, complicated the situation further (context). In some cases, conversations about dying, including ACP conversations, were considered to be the remit of the palliative care team. There was a lack of shared ownership between the teams with regard to these elements of the child's care.

Organisational culture was a very significant influence. The biomedically orientated culture in healthcare reinforced the implementation of guidelines and protocols, the need to seek clinical certainty (and avoid uncertainty), onward referral for new clinical concerns and the avoidance of the

possibility of death. Clinicians working within this culture were conditioned to maintain the focus on cure (mechanism), which was further reinforced by the situation that the death of a child was an abhorrent prospect (mechanism). These contexts and outcomes perpetuated each other. The death-defying and death-denying culture (context) constrained the delivery of palliative care, including referrals to specialist palliative care services (outcome). The contexts, mechanisms and outcomes leading to a collusion of immortality are summarised in figure 9.4.2:

Figure 9.4.2: CMOC 8 - The collusion of immortality
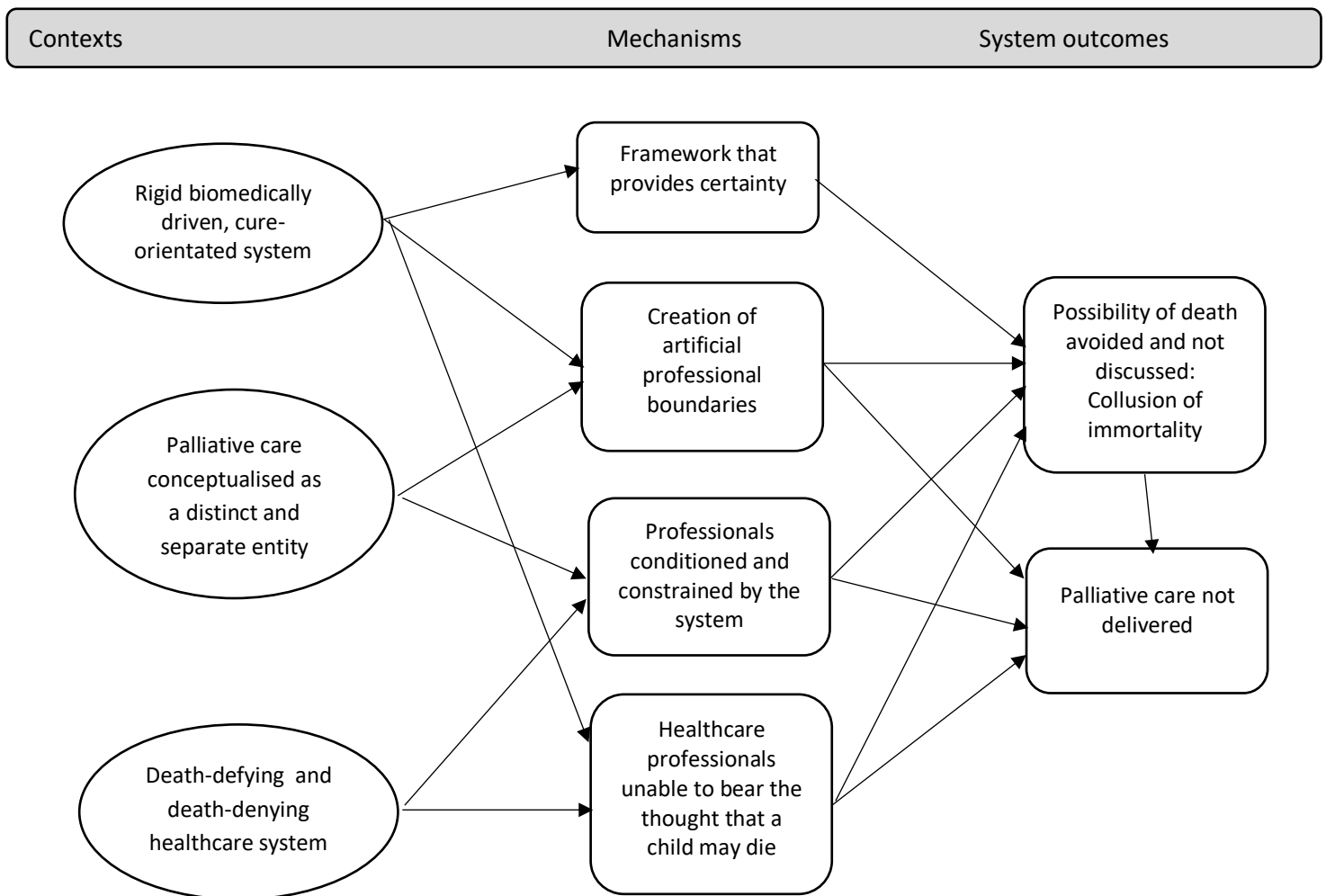
CMOC 9: Delivering a palliative care counterbalance
CMOCs 7 and 8 provide detailed insights from the empirical research data, into how the healthcare system can affect the experiences of children living with life-limiting and life-threatening conditions and their families. The CMOCs propose how and why in this healthcare system palliative care is inconsistent, including referral to specialist services. They also provide insights into child and family relevant outcomes, and a detailed basis from which to consider how the situation can be improved.

Palliative care was conceptualised inconsistently by both family members and healthcare professionals. Palliative care professionals advocated for palliative care as a broad approach to care. However, it tended to be conceptualised as a distinct entity, viewed as a distinct specialty or service, which added to the fragmentation of the healthcare system. A significant barrier described by families was an association between the word "palliative" and "giving up" or "dying". The children who were aware of palliative care services also regarded the term negatively. Where professionals felt that a child and family could benefit from a palliative care service, referrals were sometimes made without using the term "palliative" in discussions with families. In the focus groups, healthcare professionals described palliative care as a "blind spot" amongst their colleagues in other specialties. Some were critical of their colleagues' delivery of palliative care, raising concerns about prescribing at the end of life and patient safety. There were tensions between the delivery of palliative care as an approach to care, and palliative care as a specialist service delivered within a finite resource. There was also recognition that multiple system factors may be prohibiting the delivery of palliative care by non-specialist colleagues, including a lack of time, continuity and the collusion of immortality (as outlined in previous CMOCs).

This CMOC aims to draw upon the research findings and previous CMOCs to propose strategies to overcome micro, meso and macro-system barriers and provide palliative care to children and their families. Throughout this CMOC, contexts are referred to at micro, meso and macro-system levels, each triggering certain mechanisms to achieve child and family outcomes relevant to the delivery of effective palliative care. The focus of this CMOC is to bring together and describe the contexts, mechanisms and outcomes that make palliative care possible, which is an important step towards developing a programme theory from the empirical research.

Several successful strategies to improve palliative care delivery were described in the research data. Firstly, the physical presence of a specialist paediatric palliative care team (meso-system context) influenced the behaviours of others through role-modelling and legitimising a palliative approach to care (mechanisms). The specialist team became allies to other healthcare professionals (mechanism), and could support them through emotionally charged conversations with families (outcome). Integration into other specialist teams, by attending clinical team meetings (meso-system context), and becoming accepted as a member of the team in those meetings (mechanism), slowly had an impact on the behaviour of other groups of professionals in terms of being more able to consider palliative care as an important part of the child's management plan (outcome). Finally, agreeing critical moments within a child's illness and developing a referral protocol to palliative care seemed effective (meso-system context). Presenting a referral to palliative care services (outcome)

as a routine and normal part of care (mechanism) at, for example, the time of relapse for a child with an oncology diagnosis, was described as a successful strategy. In this situation, it helped that the clinicians working in oncology were particularly interested in palliative care and also worked in the local children's hospice (meso-system context). The same professional providing care throughout a child's life having an interest in palliative care (micro-system context) and continuing to care for the child as the end of life approached was also described as effective (outcome).

The benefits described by families who had palliative care teams involved in their child's healthcare often revolved around the behaviours of individual healthcare professionals who worked in these services (micro-system context). They advocated for the children and families in difficult situations and assisted with the co-ordination of the child's care (outcomes). For some, the palliative care team provided a particular healthcare intervention (for example a regular injection in the community for C013) which no other team was willing to deliver. Families would regularly refer to palliative care professionals as those who they turned to when circumstances were particularly difficult, such as when their child was very unwell, or when they had concerns that no other specialist team had addressed (outcome). The palliative care team validated their concerns (mechanism) and took action to ensure that their care needs were met (outcome). One family suggested renaming the service the "sunshine service", since the approach to care that they received from individuals in this service provided them with a feeling of enhanced wellbeing because they felt cared for (outcome).

The delivery of palliative care, including referrals to specialist paediatric palliative care services, depended on individual healthcare professionals who were motivated in palliative care or who were able to acknowledge that the death of the child was possible (micro-system context). These individuals acted in ways that challenged the collusion of immortality (mechanism). The approach provided by these professionals was holistic, and included validating the concerns of the child and family, addressing the existential, spiritual and psychological needs of the child and family, and acknowledging death as a possibility (outcome).

One of the many challenges that the professionals who took part in the focus groups described as being a barrier to the delivery of palliative care was that "there's nothing inspirational about palliative care". At a macro-system level, the potential impact of senior leaders of healthcare organisations who prioritised palliative care was very clear (macro-system context). Individual system leaders who recognised the importance and potential of palliative care, and championed the development of specialist paediatric palliative care (mechanisms), had a key role in the development of services, including investment where possible. The presence of these leaders, and subsequent investment in and development of services, was associated with a cultural shift at an organisational

level (mechanism) which further legitimised and supported the delivery of palliative care (outcome). If those individual leaders moved on, there was a risk that this cultural shift would be undone.

Figure 9.4.3 provides a summary diagram of these micro, meso and macro level contexts, mechanisms and outcomes that together enable the delivery of palliative care:

Figure 9.4.3: CMOC 9 - Delivering a palliative care counterbalance
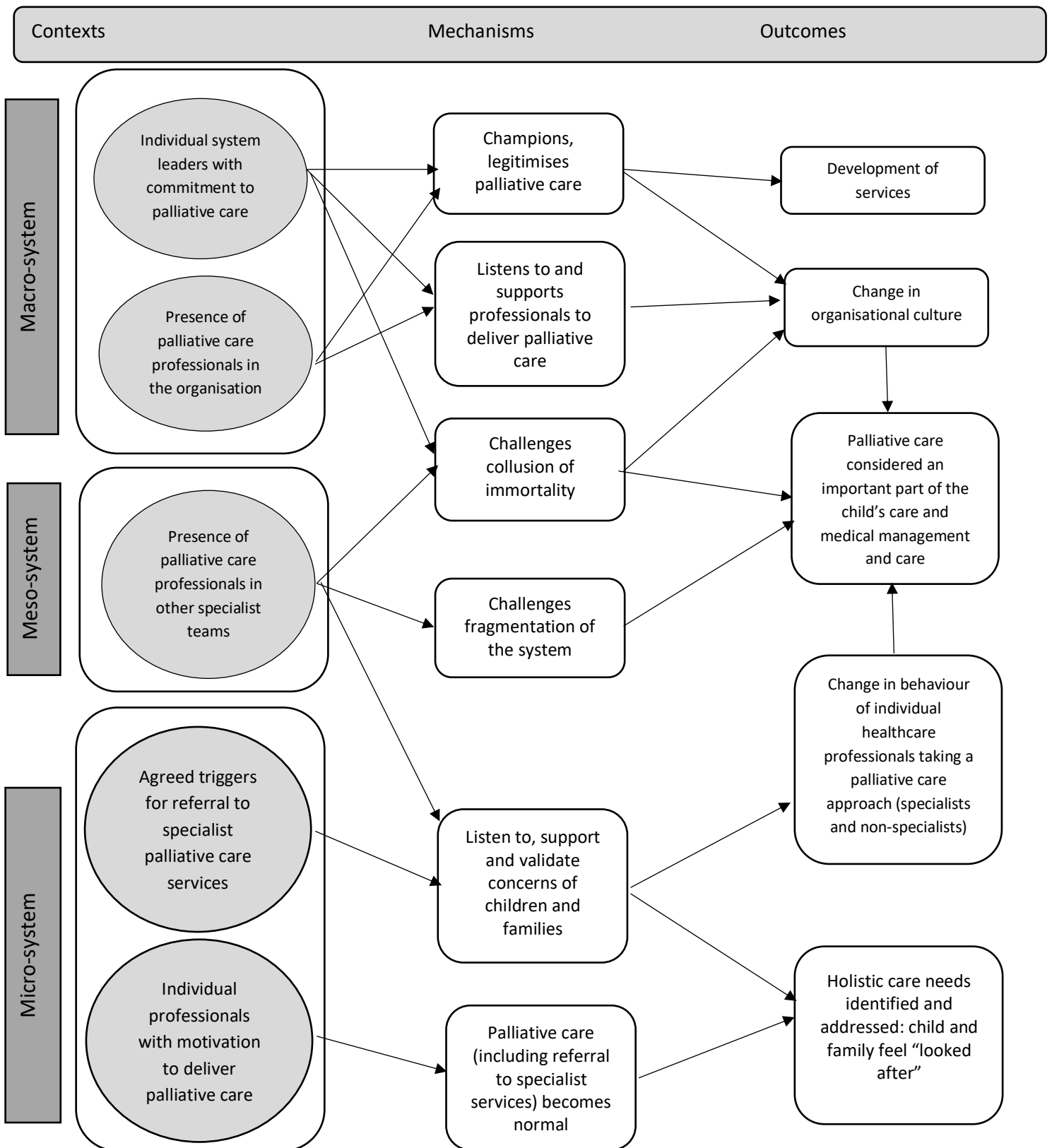
9.5. Chapter summary

The CMOCs outlined through the application of a realist logic to the findings of the thematic analysis are not exhaustive but describe important contexts and outcomes, drawn from the empirical data, which require consideration in the delivery of palliative care (both as a broad approach and as a specialist service). The mechanisms abstracted from the data are triggered in the contexts described, to produce the outcomes. The CMOCs have been configured to show in which contexts certain mechanisms are triggered, leading to the outcomes. These detailed findings will be used to inform the development of a programme theory in chapter 10.

Part Four: The Programme Theory, Discussion and Recommendations

10.   Development of the programme theory

10.1.   Overview of Chapter 10

This chapter describes the development of the programme theory, through situating the findings of the empirical research within the wider literature, as described in the literature reviews (Chapter 3). The programme theory proposed as a result of the literature reviews has been tested, refined and refuted with the findings of the empirical research. The steps taken in order to develop the programme theory (as outlined in the Chapter 5, Methods) are:

1. Identification of an initial programme theory that palliative care for children "works" from policy statements and the systematic review.

2. Testing and further development of the programme theory through a realist literature review to identify the CMOCs that provide insights into how palliative care for children "works", and in what circumstances

3. A process of refining and refuting the CMOCs from the realist review with findings and CMOCs from the empirical research, to understand how, when, in what circumstances and why palliative care "works" for children and their families.

The focus of the analysis and theory development is generative causation, providing explanations about how and why child and family outcomes can be achieved, to lead to policy-relevant recommendations for the future delivery of palliative care to children.

10.2.   Summary of the Realist Review Programme Theory

The systematic review (Chapter 3, Section 3) provided limited evidence that specialist paediatric palliative care services "work" for children and families, with beneficial outcomes including a feeling of support for families, improved quality of life, and achieving a preferred place of care. The realist review (Chapter 3, Section 4) investigated how palliative care provides benefit to children, when, how and in what circumstances.

The programme theory from the realist review proposed important child and family-related outcomes that underpin the delivery of palliative care, including feeling respected, heard and supported, and having an ability to place the emphasis of care on lessening the child's suffering (outcomes). These outcomes depended on established, trusted relationships with healthcare professionals. An ability to develop these relationships through consistency and continuity of care, and motivation to deliver palliative care, were necessary amongst professionals (contexts). The

mechanisms that underpinned these relationships included respect for the family circumstances, advocacy, affirmation, an ability in the healthcare professional to bear witness to the child and family situation, and emotional investment in the relationship. Through these relationships, shared emotional impact and open acknowledgement of the fragility of the child's condition and the possibility of dying could be achieved (outcomes). These outcomes were proposed as key precursors to conversations during which child and family preferences and priorities, and referral to specialist paediatric palliative care services, could be discussed. It was proposed that achieving these outcomes could support more consistent delivery of the service outcomes identified in the systematic review, including improved quality of life and symptom control, and a feeling of support for families. In turn, policy outcomes, including the formulation of an Advance Care Plan (ACP) or achieving a preferred place of death, may also be more likely to be achieved.

10.3.   Development of the programme theory

The literature reviews focussed on palliative care delivery. The empirical research focussed on the healthcare experiences of children with life-limiting and life-threatening conditions, and how they relate to palliative care, or not. Insights and understanding from the empirical research have been used to test and develop a new programme theory to inform the delivery of palliative care for children in the future.

The following questions were used to guide the development of the programme theory:

- What are the most important outcomes from the child and family perspective? How have they been achieved?
- What causal mechanisms have operated?
- What are the contexts that trigger these causal mechanisms, in what contexts have the mechanisms been triggered to produce these outcomes?
- How do the micro, meso and macro-system CMOCs interrelate?
- What is necessary in order for palliative care, as a complex intervention, to "work"?

The programme theory development has focussed on contexts, mechanisms and outcomes at micro, meso and macro-system levels, and the relationships between them, with the specific aim of informing the future delivery of palliative care for children. Drawing together the CMOCs from the literature review and the empirical research results in frequent overlap and repetition. There are several examples of outcomes from one CMOC becoming contexts for another, including for example, the family having expert knowledge of their child's condition and care, the child's condition becoming a vocation (outcomes in CMOC 1), which both function as contexts within which palliative

care is delivered. This chapter describes the systematic approach taken to the development of the programme theory.

1. *The child and family as vulnerable experts*

The development of the programme theory began by considering the child and family contexts. Both the realist review and the empirical research provided important insights into the child and family contexts in which palliative care must be delivered.

Every child and family situation was highly individual, in terms of both the child's condition and the family circumstances. There were immense and intense emotional burdens for families when a child developed a life-limiting or life-threatening condition. This completely disrupted the family narrative, changing life forever (CMOC 1) (28, 144, 157, 164, 174). The child's condition was an imposition, not a situation of choice, and there were immediate obligations for family members. They embarked on a continual process of adaptation and learning to cope with their child's condition. This required managing not only the emotional impact on themselves, changes in their personal identity and their hopes and expectations for parenthood (CMOC 1) (142, 144, 153, 157, 164, 168, 169), but also the practical challenges associated with the condition and with managing the healthcare system (CMOCs 1-3) (172). Families drew upon a variety of sources for support, including other parents and families (153, 156, 168, 178).

For many, serious and critical life-threatening deteriorations in their child's condition, followed by a period of recovery, were a frequent occurrence and shaped their hopes and expectations (CMOC 1). A new kind of normal family life occurred, where children and families were experts, and the management of the child's condition became a family vocation (CMOC 2) (27, 139, 157, 159). They expressed a desire to be respected as experts in the child's condition (134, 149, 163, 165, 166, 175, 177, 179). Significant changes occurred at home, which became a healthcare system in its own right (CMOC 2). Family members developed advanced coping strategies, and a fragile resilience in order to manage (147, 148, 154, 157, 160).

The empirical research added insights into the perspectives of children on their condition, and their relationships with healthcare professionals (CMOCs 3-4). The children who took part in the study seemed to accept their conditions as part of life. They did not want to be defined by their conditions, despite how dominant their healthcare and treatments could become, and had other priorities for life including seeing friends and going to school. This is in keeping with the findings of previous studies, where children have expressed a desire to live their lives as normally as possible despite their abnormal circumstances (33, 147, 170, 262).

In their relationships with healthcare, children valued feeling that their family members were "looked after" and that their needs were being met (CMOC 4-5). Important child and family outcomes in the both the literature and the empirical research were feeling respected, heard and supported by their healthcare professionals (134, 149, 163, 165, 166, 174, 179).

Most of the children who took part in this study had some understanding of the life-threatening nature of their condition, and that death was possible, regardless of their age (CMOC 6). They trusted the adults around them, and were usually passive recipients of the decisions made in relation to their health and care (115). The children wanted their ideas and priorities to be heard, but also wanted to protect and please the adults around them, a situation that could cause cognitive dissonance and affected their psychological wellbeing (CMOC 3)(137).

The families who took part in the study all had an awareness of the life-threatening nature of their child's condition. They all knew that the death of their child was possible, but this tended to be alluded to rather than spoken about openly (CMOC 6). As in previous studies, life with intensive medical treatments and chronic uncertainty became normal (27, 139, 157, 159). Parents have anxieties about a right or wrong way to discuss death and dying with their children (137, 144, 171, 174). They describe seeing their child's understanding of their situation change over time (139), with a "tacit understanding" that they may die developing, such that explicit conversations about dying become unnecessary (29, 174).

The programme theory therefore begins with the context of the child and family as vulnerable experts. The detailed aspects of the CMOCs related to the child and family situation are not detailed again in the programme theory, so this context is multi-layered, as in figure 10.1:

Figure 10.1: Child and family contexts at the centre of the Programme Theory:

2. *The child and family within the healthcare system*

By the nature of the child's condition, the child and their family had to function within and manage a complex healthcare system, which they experienced at micro (interpersonal), meso (organisationa) and macro (cultural) system levels. Palliative care must be delivered within this complex healthcare system, which has previously been described as both "bewildering" (51), and "disempowering" (115, 160, 170). Figure 10.2 shows the child and family as vulnerable experts (context) at the centre of a complex healthcare system, with the outcomes they described as important: feeling "looked after" and respected.

Figure 10.2: The child and family within the healthcare system
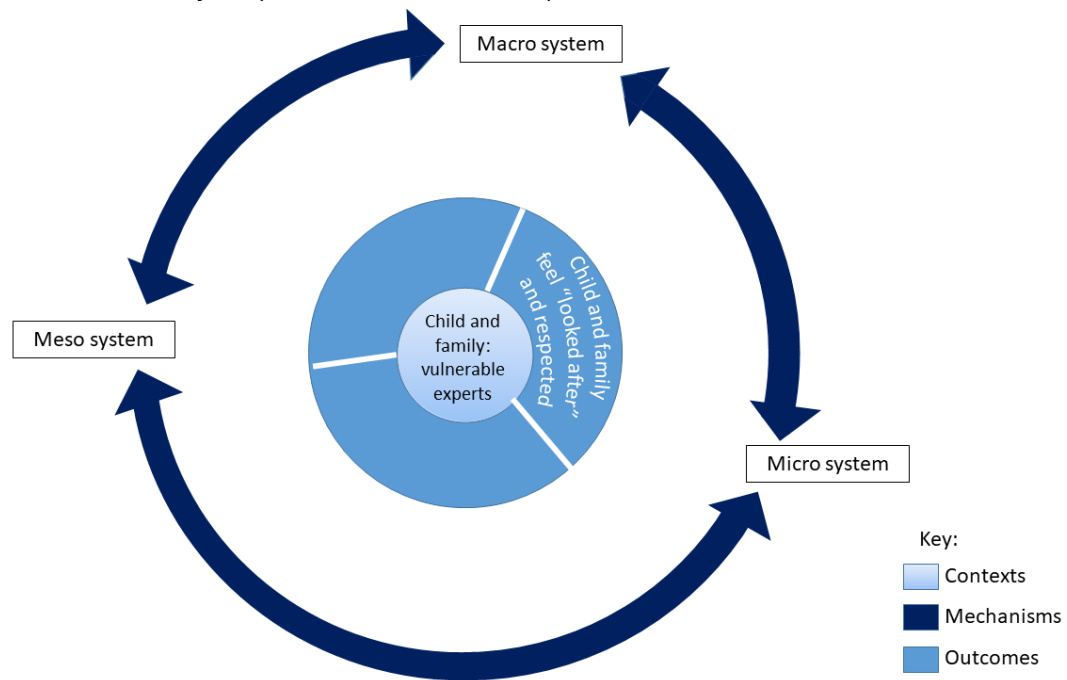
3. *Relationships with professionals key micro and meso-system contexts*

The child and family interactions with healthcare services at the interpersonal (micro-system) level were key to their experiences of healthcare, including palliative care (CMOCs 4 and 5) (92, 119, 131). Trusted, authentic relationships, in which professionals could develop detailed knowledge of the child and their family, were highly valued, and were crucial to the child and family feeling "looked after" and secure. A combination of factors contributed to achieving those outcomes. Consistency of healthcare professional either in a particular healthcare environment, or delivered through relational continuity of care (CMOC 4) (33, 170) played a part. The motivation of individual professionals to provide a compassionate, individualised approach to care was also important. Open and honest communication, care co-ordination, accessibility and availability were all valued (CMOCs 4 and 5) (33, 146, 151, 160, 162, 173, 179, 181). The mechanisms which led to these outcomes included respect for the family circumstances, advocacy, and affirmation in decision-making, a

198

capacity in the healthcare professional to bear witness to the child and family situation, and emotional investment in the relationship (CMOCs 4 and 5)(51, 98, 164, 171, 173).

Through these relationships, shared emotional impact, open acknowledgement of the fragility of the child's condition and the possibility of dying could sometimes be achieved. Mechanisms which were triggered in these contexts included advocacy and sharing the emotional burden (CMOCs 4 and 5). These established, trusted relationships were often experienced with professionals working as part of a palliative care service, including children's palliative care community nurses, but the approach was experienced in child and family relationships with a wide range of other professionals too, for example, GPs, surgeons, oncologists, specialist paediatricians, dieticians and specialist pain nurses. It was recognised by both family members and healthcare professionals who took part in this study that the development of these relationships caused vulnerability (mechanism) amongst healthcare professionals (CMOC 5), and that there was a need to ensure support for healthcare professionals as well as children and families. These are important factors in enabling the contexts in which micro-system relationships can be developed, resulting in children and families feeling respected, heard and supported.

Figure 10.3 adds connections with healthcare professionals, enabled through consistency, continuity and the motivation and values of the healthcare professional, to the developing programme theory as a key context for the development of trusted interpersonal relationships, and for the delivery of palliative care.

Figure 10.3: Relationships with professionals as a key context
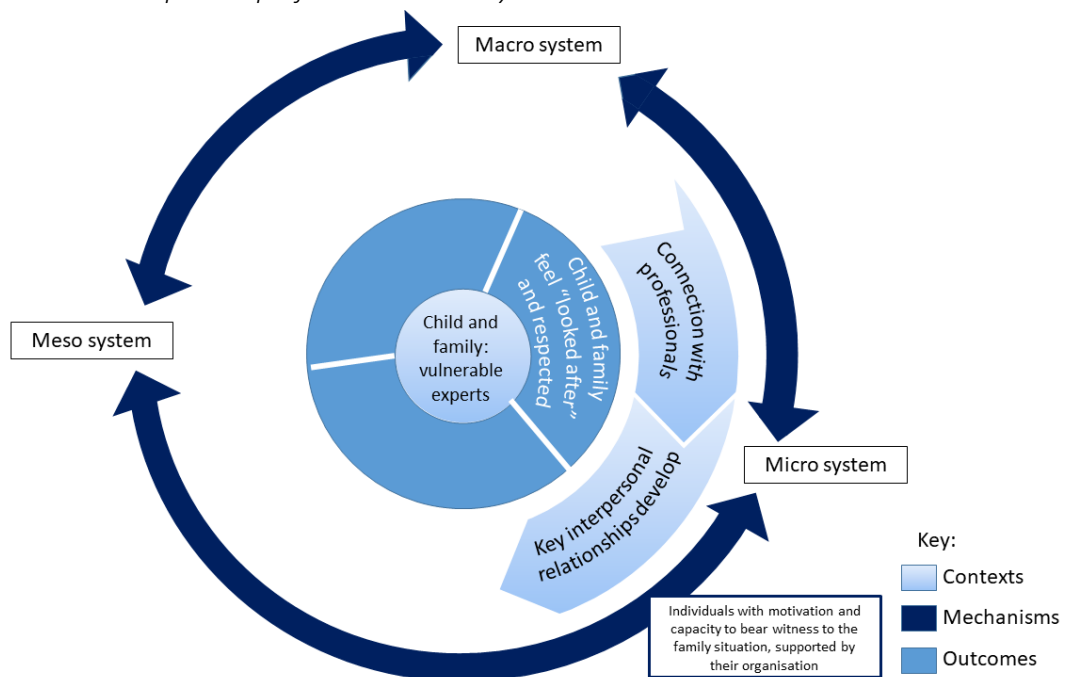
4. Challenging the "collusion of immortality" at the meso and micro-system levels

Children and family members experienced a lack of continuity both within individual specialties, and across the healthcare system. Every family described "fighting" a rigid, fragmented system, in order to obtain the care that their child needed (CMOC 7) (33). A lack of co-ordination of care between different specialties could be "disturbing" for families (135). Communication between specialities and opportunities for the different specialists involved in the care of the child to come together with the family were limited. Children and families entered into a "collusion of immortality", created within a death-defying system and with multiple opportunities for the possibility of death to be avoided in discussion with healthcare professionals (CMOC 8). This did not necessarily take into account the level of uncertainty or complexity related to the child's condition, nor did it allow for exploration of the child or family's knowledge that the child may die, nor did it openly address these concerns with families (CMOCs 7-8). Within this system, interpersonal relationships with individual healthcare professionals who shared their emotional burden and advocated for them, became even more important. It was within these interpersonal relationships that uncertainty and the possibility that the child may die could be acknowledged. These were key precursors to conversations during which child and family preferences and priorities, and referral to specialist paediatric palliative care services, could be discussed (CMOC 9).

Palliative care was frequently conceptualised as a distinct service and often associated with the end of life. The "p word" problem described by the children, families and healthcare professionals in this study was a perceived barrier to referral to palliative care services. Previous studies have found referrals to palliative care to be associated with fear amongst families that they would lose contact with the healthcare professionals and services they knew and were familiar with (180). The systematic review (chapter 3) showed a range of benefits associated with care from specialist paediatric palliative care services including a feeling of support for families and improved symptom control (96-106). Symptom control could be particularly challenging given each child's individual condition and circumstances (39, 96, 115, 143, 158), requiring specialist expertise.

At the micro-system level, and from the perspective of children and families in this study, paediatric palliative care teams attended to a variety care needs (CMOC 9). For some children, the palliative care team delivered interventions that no other team was willing to provide. Palliative care team members were often involved in advocating for families and co-ordinating aspects of their care. However, palliative care teams were not solely responsible for these aspects of care, and did not necessarily consider this to be their key role or function. Specialists in paediatric palliative medicine who took part in the focus groups emphasised their role in complex symptom management,

involvement in discussions about limitation of treatment ("ceilings of care") and holistic support for families.

At the meso-system level, the presence of a specialist paediatric palliative care team within an organisation had an impact beyond the provision of hands-on care to children. There was evidence in the empirical data that the presence of the team also influenced the behaviour of other healthcare professionals, through mechanisms including role-modelling and legitimising the palliative care approach; illuminating the "blind spot" (CMOC 9) (195).

A specialist paediatric palliative care team has been added to the emerging programme theory outlined in figure 10.4. The proposed theory of how palliative care delivery could be improved states that access to specialist services depends not only on their presence, but on the presence of other contexts, including key interpersonal relationships between children, family members and healthcare professionals. These depend on consistency and a commitment amongst those professionals to a holistic approach to care, with acknowledgement of uncertainty and the possibility of the child dying, whether this is spoken or not. Individual healthcare professionals are required to act in a way that challenges the collusion of immortality. Mechanisms triggered by the presence of the specialist paediatric palliative care team help, including role-modelling, legitimisation of palliative care and culture change. These are important influences in changing attitudes and culture within a healthcare organisation, supporting and enabling professionals from other specialties to deliver a palliative care approach (as in CMOCs 4-5):

Figure 10.4: Challenging the collusion of immortality
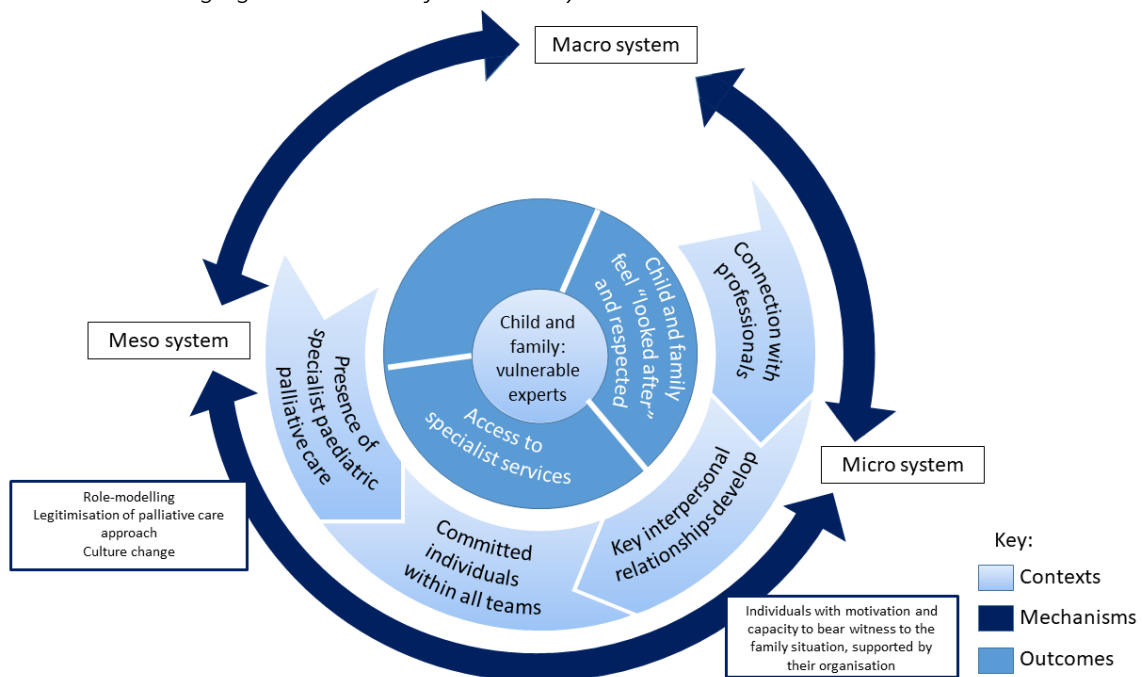
5. Committed leadership as a key macro-system context

The potential impact of committed organisational system leadership was highlighted during the focus groups. Participants described a need for culture change in healthcare organisations, with a focus on joined up holistic care, co-ordination of services, keeping the patient and family at the centre feeling respected and cared for. These aspects of care were not considered particular to palliative care services, but were described as necessary across the system if palliative care was to be delivered to children and their families.

Focus group participants described the key role that healthcare leaders had taken in the development of specialist paediatric palliative care services in the past. As well as finding financial resource to support the development of services, their approach and commitment also challenged the collusion of immortality (CMOC 9). Over time, this approach had the potential not only to improve access to specialist services (outcome), but also to reinforce the value of palliative care within the organisation and change culture (contexts). Mechanisms included role-modelling and legitimising palliative care. These have been added to the programme theory diagram in figure 10.5 below.

Changes in leadership were also relevant. New leaders, who focussed on different organisational agendas, were associated with disruption to this process of culture change.

Figure10.5: The potential impact of committed organisational leadership (a macro-system context)
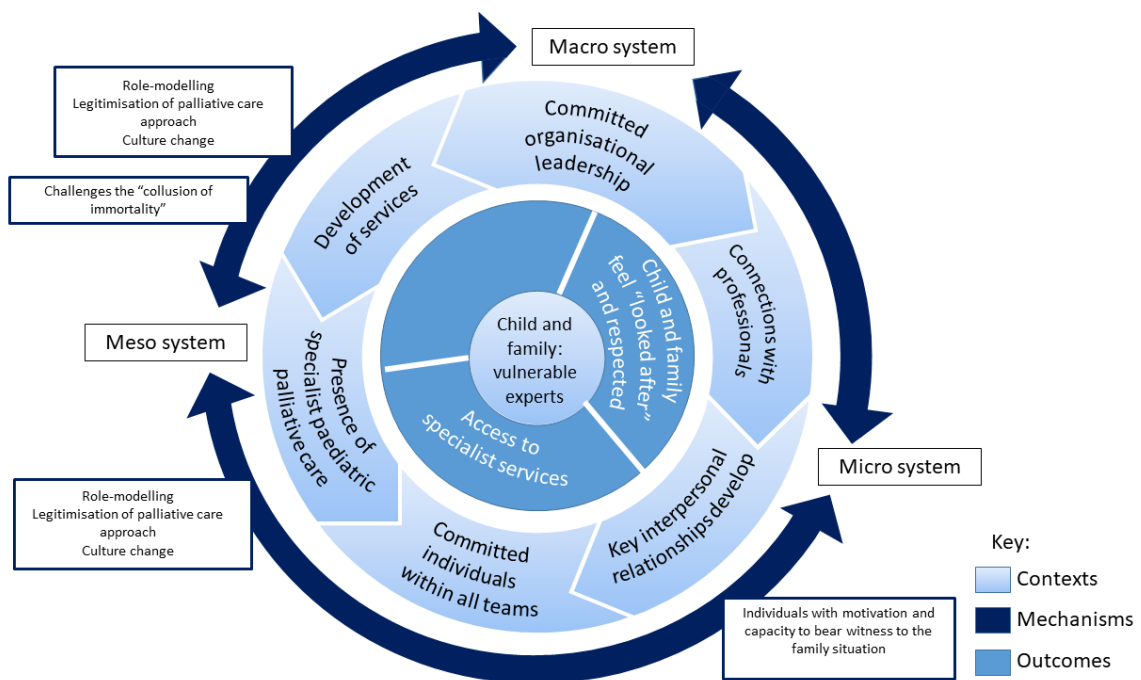
10.4.   Summary and consolidation into an overarching programme theory

The overall programme theory outlined in figure 10.6 on page 204 shows how complex factors related to the delivery of palliative care at the micro, meso and macro-level interrelate. The child and family are held firmly at the centre of the programme theory, described as "vulnerable experts". Interpersonal relationships with healthcare professionals at the micro-system level are key. Organisational culture that places emphasis on the provision of well co-ordinated, holistic care, and that enables the development of relationships between children, families and healthcare professionals is important. In these contexts, mechanisms are triggered which lead to children and their families feel heard and supported as an outcome, and this provides a basis for the delivery of palliative care as a broad approach by both specialists and non-specialists. Figure 10.6 provides a diagrammatic presentation of the proposed overarching programme theory for the delivery of palliative care to children that has resulted from this research. The mechanisms that bind together the micro, meso and macro-level contexts to produce the child and family related outcomes are presented in relation to one another. How these may relate to achieving policy outcomes, and the underlying mechanisms that are necessary, have also been added to the diagram in purple:

Figure 10.6: A programme theory for the future delivery of palliative care to children and families

Role-modelling
Legitimisation of palliative care approach
Culture change

Recognition of palliative care as a priority, agreed and understood, challenges the "collusion of immortality" and changes culture

Macro system

Committed organisational leadership

Development of services

Values compassion and continuity and supports interpersonal relationships between children, families and healthcare professionals

Connections with professionals

Meso system

Presence of specialist paediatric palliative care

Policy outcomes achieved

Child and family: vulnerable experts

Child and family feel "looked after" and respected

Access to specialist services

Micro system

Role-modelling
Legitimisation of palliative care approach
Culture change

Embedding the approach across the system to decrease fragmentation

Committed individuals within all teams

Key interpersonal relationships develop

Individuals with motivation and capacity to bear witness to the family situation, supported by their organisation

Key:

Contexts

Mechanisms

Outcomes

11. Discussion

11.1. Overview of Chapter 11

The programme theory development in Chapter 10 brought together the findings of the empirical research with the findings of the literature reviews. In this chapter, there is a description of the contribution of the research to knowledge, how the research has met its objectives and how the research questions have been addressed. The methodological considerations, strengths and limitations of the research are discussed, followed by a set of recommendations for policy and practice, which have been drawn from the programme theory proposed in Chapter 10.

11.2. Discussion of the research findings

The research has provided in-depth insights into the experiences and perceptions of healthcare of children with life-limiting and life-threatening conditions and their families. The research makes an important contribution to knowledge firstly in that it has captured the views of children. Secondly, through the realist approach, the research has provided a description of outcomes most important to children and families in the delivery of their healthcare when the child has a life-limiting or life-threatening condition, and thirdly, the research has led to the generation of a new programme theory for the delivery of palliative care to children. The proposed programme theory has informed the development of policy-relevant recommendations outlined later in this chapter.

Complexity theory underpins the research. Complexity theory recognises that clinical practice, organisation, information, management, research, education and professional development are interdependent and delivered through multiple self-adjusting and interacting systems, with constant uncertainty and unpredictability within the system (228). The sources of complexity in healthcare, and palliative care, for children with life-limiting and life-threatening conditions, are multiple. They include the highly complex, individual, unpredictable conditions that the children live with, the impact on the family, and the dynamic and sometimes unpredictable organisation of the healthcare system. Complexity theory goes further to describe healthcare systems as dynamic, characterised by uncertainty, unpredictability and emergence, all of which are relevant to the future delivery of palliative care for children (263). There have been calls for a paradigm shift away from traditional "reduce and resolve" approaches to clinical care and service organisation in order to improve healthcare delivery to an increasingly complex population (229). Complexity science provides a useful framework to consider how palliative care, as a complex intervention, could be delivered to the diverse and increasing population of children with life-limiting and life-threatening conditions, and may be more useful than other, more widely accepted models for innovation and change, such as implementation science (263).

One of the most important contributions of the research is that it has captured the views of children, along with those of their families and healthcare professionals involved in the delivery of palliative care for children. The literature reviews revealed a lack of previous research in this area where children have been participants.

A further contribution of the research is the realist approach to the study of palliative care, as a broad approach to care for children with life-limiting and life-threatening conditions, as outlined in widely accepted definitions (1, 2). The realist approach is a systematic, explanatory approach, appropriate for the study of multi-faceted, complex interventions. This study provides increased understanding into the experiences of healthcare of children with life-limiting and life-threatening conditions and their families, how they conceptualise palliative care, how this affects their experiences, and why, when and how a palliative care approach "works". Realist analysis of the data from the serial interviews with children and their families and healthcare professional focus groups, along with the insights provided by the literature reviews, has provided an understanding of the hidden explanatory mechanisms that underpin the delivery of palliative care for children, and informed the programme theory. The programme theory provides a dynamic framework that acknowledges complexity at micro, meso and macro-system levels, and forms the basis of recommendations for future policy, outlined later in this chapter. The realist approach also allows for increased recognition of the contexts that are required to activate mechanisms in order to achieve desirable child and family outcomes, and has the potential to assist the translation of policy into practice.

The following section of the chapter is a discussion of the key research findings, how the research has addressed each of the research questions, and what the research adds to current knowledge.

Research question 1: How do current definitions of "palliative care" for children concord with service delivery, policy and guidance in the UK?
The term "palliative care" is associated with a wide range of views amongst children, families and healthcare professionals, and is used inconsistently in policy and practice. Patient and public involvement (PPI) work and the research findings both suggested that current definitions of "palliative care" for children concord poorly with service delivery, policy and guidance in the UK.

PPI early in the course of the research highlighted some of the diverse views and opinions that exist around the term "palliative care". Specifically, young people who had received hospice care, or whose siblings had experience of palliative care services, were not comfortable with the term "palliative care", and felt that it carried negative connotations. One young person described palliative care as "a distant and all-encompassing concept that does not drive understanding" (264).

Similar concerns about the negative connotations of the word "palliative" were borne out throughout through the literature reviews, and the data collection. The realist review identified variable perceptions and opinions of the term "palliative care" amongst professionals (18), children, and their families (124, 146), as a barrier to referral to specialist services. Family members viewed "palliative care" as a distinct phase at the end of a child's life, "the beginning of the end", and feared it as a point at which they would lose contact with the healthcare services they knew, a situation that was considered to be "terrifying" (180).

Every child who took part in the empirical research study had a life-limiting or life-threatening condition and palliative care needs, according to current categories and definitions, but less than half of them (6/14) had a palliative care team involved in their care at the time of the interviews. Whether or not they were under the care of a palliative care team, the term "palliative" had negative connotations for the children and their families, with the "p word" problem emerging as a key theme during the data analysis. "Palliative care" tended to refer to a particular healthcare professional or service, or a phase in a child's condition, rather than a broad philosophy of care aimed at improving quality of life. Furthermore, the research found marked inconsistencies in the services delivered by palliative care teams. When they were involved in the care of the children, palliative care teams were often those called upon by families to address gaps in care such as care co-ordination, the delivery of certain interventions in the community, and specific symptom control issues.

Research question 2: What is the current evidence base for practice and policy related to palliative care service delivery for children?
The literature reviews provided a detailed overview of the current evidence base in children's palliative care for practice and policy. The literature reviews highlighted a paucity of research evidence in children's palliative care. Many published articles in children's palliative care are expert opinion, rather than research papers. The research papers identified during the literature reviews were heterogeneous in terms of research design. In particular, there was a lack of research related to the child's experience, with only seven studies including children as participants (33, 154, 158, 162, 167, 170), five of which involved interviews with children, with or without their parents (33, 154, 162, 167, 170). No longitudinal studies that investigated the changing family experience over time were identified in the review.

The systematic review provided evidence that specialist paediatric palliative care services, as defined in current guidelines as those supported by a consultant in paediatric palliative medicine, provide benefits to children and families. Benefits included a feeling of support, improved quality of life

including symptom control and activities that brought meaning to the children's lives, and an increased likelihood of achieving a preferred place of care and death. The published evidence base was small and heterogeneous in terms of study design and quality, but supported current policy recommendations for the involvement of specialist paediatric palliative care services in the healthcare of children with life-limiting and life-threatening conditions and their families. The review informed the initial programme theory that palliative care for children "works", but provided little insight into *how* specialist paediatric palliative care services achieved beneficial outcomes for children and families.

The focus of the realist review was the child and family experience of palliative care. Published research where the participants were children and family members, and personal accounts from family members, were included in the review. Context-mechanism-outcome configurations (CMOCs) were extrapolated from the data to describe the hidden mechanisms that were triggered in certain contexts in order to produce desirable outcomes for children and families, building theory to propose how palliative care works, for whom and in what circumstances.

The programme theory from the realist review proposed two interdependent contexts: the family situation (the family as experts, and the child with their own priorities), and professional resource contexts (established and trusted relationships between healthcare professionals, children and their families). In these contexts, mechanisms were triggered, including advocacy and affirmation in decision-making, a capacity amongst healthcare professionals to bear witness to the child and family situation and emotional investment. These led to important child and family outcomes that underpin palliative care delivery, including referrals to specialist paediatric palliative care services. The review found that important child and family outcomes included feeling respected, heard and supported, having an ability to share the emotional impact of the child's condition, placing an emphasis of care on lessening suffering, and acknowledging (sometimes implicitly) the possibility of death.

Research question 3: What are the lived experiences of children with life-limiting and life-threatening conditions and their families?
The literature reviews provided insights into the lived experiences of children with life-limiting and life-threatening conditions, and their families. The empirical research added to these insights, through serial interviews that included the views and experiences of the children. Focus groups with healthcare professionals involved in the delivery of healthcare, including palliative care, to children and families, provided further opportunity to explore the findings from the interviews and triangulate them with the professional perspective.

This research found that the children lived in the present, had their own interests and priorities (beyond healthcare), and seemed to accept their condition as part of life. They did not wish to be defined by their condition and, in keeping with previous studies, had a desire to maintain normal life as much as possible given their highly abnormal circumstances (33, 262).

The children had in-depth knowledge of their conditions, and experienced a range of different symptoms, but did not tend to talk about these during the interviews. A review published in 2019 described the multidimensional, complex symptoms experienced by children with both malignant and non-malignant conditions, with overlap between the two groups, and highlighted a need to improve the holistic assessment and management of these symptoms, including the psychological elements that were contributing (265). Family members in this study reported their children experiencing both physical and psychological distress, and described a perception that healthcare professionals placed greater emphasis on managing physical symptoms, often with medication or an onward referral to another specialist.

When a child was born with, or developed a life-limiting or life-threatening condition, it changed family life forever. In keeping with previous research, this research found that family members, usually parents, were obliged to take on a new role as carer (28, 266), which had an impact not only on everyday life, but also on their sense of purpose and identity. Over time, the child's condition became a vocation and the home became a healthcare system in its own right. Family members became experts in the management of their child and their child's condition, negotiating the various systems, including healthcare, that were involved in the provision of their care. They developed sophisticated coping strategies, and a vulnerable resilience, in order to deal with the fluctuating trajectory and fragility of their child's situation, which, over time, became normal. The situation that the families found themselves in had not arisen through choice. This is an important point to remember when there is emphasis on choice in the rhetoric of healthcare policy in palliative and end of life care (267).

Research question 4: How do children with life-limiting and life-threatening conditions and their family members perceive healthcare services, including "palliative care"?
The healthcare experiences of children and families were mixed. Those who took part in the study expressed a desire for proactive, holistic, well co-ordinated healthcare, which seemed to be lacking. Palliative care tended to be viewed as a specialist service by the children and their families. There was variation in terms of both the availability of palliative care teams, and the roles they played in the delivery of care. The programme theory proposes that the provision of palliative care, as a broad approach to care, including referral to a specialist paediatric palliative care service where possible,

depends on the delivery of the proactive, holistic, well co-ordinated healthcare that children and families desire.

At the interpersonal (micro-system) level, trusted relationships with individual healthcare professionals were critical to the child and family experience. Children valued healthcare professionals who made them feel "looked-after", and who attended to the needs of their families. They spoke about particular individuals who had displayed interest in their lives beyond healthcare, such as school or a sports team. The children valued seeing familiar professionals, who they trusted. They also expressed a desire for a healthcare professional to regularly "check-in" with them, and to attend to the needs of their family members.

Previous research suggests that children wish to be more involved in decisions about their own healthcare (268). A range of barriers to this involvement in decision-making has been described previously, including a restricted ability in children to express their needs and wishes, and communication that takes place between healthcare professionals and parents, leaving children feeling unable to participate even when they wish to (269-271). The children in this study tended to trust their adult caregivers and healthcare professionals to make decisions about their care and treatment, but at times felt unheard. Some of the children did not have the mental capacity or verbal communication to enable them to take part in decision making about their care, which provided further challenges, particularly as they became young adults and made the transition to adult healthcare services. Shuttle diplomacy, where the healthcare professional negotiates discussions between children and their family members by "shuttling" between them, has been proposed as one model of communication between children, their parents and healthcare professionals in order to navigate the most difficult and complex decisions, particularly when children do express preferences about their healthcare and management (262).

The children were perceptive to the frustrations and "fight" with the healthcare system that their family members experienced. A "fight" with the healthcare system in order to obtain the care that their child required was described by every family. For some, this was ensuring their children's symptoms were adequately managed during hospital stays, while for others the "fight" was related to elements of care such as obtaining medications, or organising many different elements of care at home. Factors that contributed to the "fight" included the fragmentation of the healthcare system, the rigidity of processes and procedures, compromised relational continuity and poorly co-ordinated care, and the "collusion of immortality". The children and their families wanted to feel listened to, and have their concerns shared and validated by their healthcare professionals. This research described the importance of professionals "listening to listen" to children and families, rather than

"listening to fix". "Listening to fix" was the approach experienced by many of the children and their families; their concerns were often addressed with the offer of a new medication or an onward referral to another specialist. This approach appeared to be more socially acceptable to healthcare professionals working in an environment heavily influenced by positivist, biomedical models of evidence-based healthcare, with rigidly followed guidelines and protocols, and an underlying death-defying and death-denying societal culture.

The importance and impact of achieving outcomes including patients feeling listened to and heard has been described in previous research. For example, Balint's concept of the "doctor as a drug" highlights the importance of healthcare consultations as clinical interventions in their own right (232). "Listening to listen" as an intervention may be undervalued in a biomedical, cure-orientated healthcare system that focusses on finding a "fix" to the problem. The outcomes that can potentially be achieved through "listening to listen" are recognised elsewhere, in other frameworks designed to place value on these nuanced aspects of consultations and care. They align with key elements outlined in descriptions of person-centred care, which include respect, coordination and integration, physical comfort and emotional support, involvement, support for the family and continuity of care (272-274). The need for increased person-centred care was highlighted by an independent inquiry commissioned by the UK Royal College of General Practitioners (RCGP) in 2014 (275). The response to the inquiry from the UK Royal College of Paediatrics and Child Health (RCPCH) called for a specific focus on patient-centred care, with new integrated care models for children and their families as well as for adults (276). Frameworks exist which are relevant and could be usefully adapted for the ongoing holistic assessment and management of the needs of these children and their families in clinical practice. One such framework is the Definitional Framework for Children with Medical Complexity, which describes the need for care and management of the condition, functional disability, family needs and healthcare use (8, 277).

Children and families valued trusted relationships. The development of these trusted relationships depended at least in part on the values and behaviours of the healthcare professionals involved, and required mutual investment. The "mutual investment fund" was described in a general practice setting by Balint as an accumulation of trust that develops between doctors and patients over many years (232). The findings of this research suggest that a similar mutual investment fund develops between children with life-limiting and life-threatening conditions, their families and healthcare professionals over days, weeks, months or years depending on the intensity of the child's condition and the amount of time spent with professionals. Within the "mutual investment fund", there was a sense of sharing the emotional impact of managing the child's condition that was key when difficult

healthcare decisions were to be made. Proposed mechanisms underpinning these relationships were an innate ability within the healthcare professional to bear witness to the child and family situation, and a personal capacity to be alongside families at difficult times. These findings align with those described in previous work as the "compassion trichotomy", which describes interdependent elements of motivation (dependent on values and personal reflection), capacity (self-awareness and regulation of energy, emotion, and cognition) and connection (a sustained patient-physician relationship) in healthcare professional compassion (278). Focus group participants emphasised the importance of their working environment, which affected their ability to provide relational continuity of care and accessibility, both of which contributed to the development of the trusted relationships that were valued by children and their families.

Continuity of care is a concept with a range of interpretations (279). Generally, continuity of care is concerned with the quality of care over time. Traditional ideas of continuity of care relate to continuous relationships between healthcare professionals and patients. As the healthcare conditions that people live with have become more complex, and the range of healthcare professionals and providers involved in the provision of their care has increased, the term "continuity of care" has also been used to describe care provided through coordination of care from multiple sources. This depends on effective sharing of information between different healthcare providers. The potential impact of the provision of relational continuity of care has been recognised in previous research (280), including as a facilitator to specific elements of palliative care for children such as advance care planning (281, 282). The findings of this research highlight the importance of relationships, with connections with healthcare providers providing a feeling of security, and the perception of compassionate care depending not only on relational continuity of care, but also on the motivation and behaviours of individual healthcare professionals.

Children and families in this research described relationships that were easily compromised by breaks in continuity, occurring if a healthcare professional was no longer available to the family due to a healthcare system change or for any personal reason. Continuity of care across services and healthcare settings was also important. A need for more effective and reliable communication between healthcare professionals across settings was identified. The implementation of effective electronic healthcare record sharing to support this continuity of care is an ongoing concern in the UK NHS. These have been extremely challenging to implement in practice, with many electronic data sharing systems (Electronic Palliative Care Coordination Systems, EPaCCS) remaining continuously "under development" (283) and have therefore not been a specific focus of this research.

The children in this study displayed acceptance of the life-threatening nature of their conditions and the possibility of death, but this was sometimes difficult for them to articulate, and was met with mixed responses from their family members and healthcare professionals. The experiences and values of the healthcare professionals involved in the care of the children affected their actions, behaviours, and the way in which they delivered care when there was a possibility that the child may die. Some healthcare professionals were able to be alongside families and acknowledge this possibility, whereas others entered into the "collusion of immortality". The "collusion of immortality" described in the research findings is also informed by Balint, who proposed that a "collusion of anonymity" exists when patients are passed around the healthcare system, from one specialist to the next, with none taking responsibility for their holistic health and wellbeing (232). The "collusion of immortality" proposed in this research describes a situation where responsibility for open discussions about the possibility of dying is passed between practitioners working within a fragmented and rigid system. The possibility of the child's death remains unspoken despite an awareness that death is a possibility amongst healthcare professionals, the child and their family members.

The "fight" with the healthcare system described by families was mirrored in the descriptions of healthcare professionals trying to deliver palliative care, who also reported a "fight". Palliative care was described as a "blind spot" amongst colleagues and "not inspirational". This fight extended to a lack of opportunity to further develop the specialty of paediatric palliative care. Paediatric palliative care services, and the professionals working within them, currently have limited capacity for the provision of frontline clinical palliative care for children. In practice, children and families received a wide variety of care and support from paediatric palliative care services, sometimes relying upon them for certain aspects of their care, including specific clinical interventions or care co-ordination that they struggled to access elsewhere. Professionals who took part in the research described palliative care as not being "inspirational" enough in a death-defying healthcare system. This was a specific challenge in gaining funding and developing new services, and caused them to feel unheard. Palliative care may lack organisational "buy-in" due to the emotional and sometimes distressing nature of contemplating the death of a child, where there is a "p word" problem, and within a death-defying and death-denying healthcare culture. Furthermore, there is recognised shortfall in the paediatric medical and nursing workforce in the UK (284). This is an area of significant concern if palliative care is not prioritised in workforce planning.

Research question 5: When and how does "palliative care" provide benefit for children with life-limiting and life-threatening conditions and their families?

This research question sought to investigate the outcomes that were most important to children and their families, the mechanisms by which these beneficial outcomes were achieved, and the contexts that determined whether or not these mechanisms produce the intended outcomes in relation to palliative care, as a broad approach to care. The programme theory outlines the elements of a healthcare system that need to be in place at the micro, meso and macro-system levels in order for important child and family outcomes to be achieved. It holds the child and family, and all of the complexities and uncertainties that they live with, firmly at the centre (context). It proposes that at an interpersonal level, established and trusted relationships with healthcare professionals (context) who are motivated to provide palliative care (context) are key. These contexts trigger underlying mechanisms including an innate ability amongst some healthcare professionals to bear witness to the child and family situation. Acts of advocacy, hearing and respecting children and their families are also mechanisms that underpin outcomes related to the delivery of palliative care. An organisational culture that values and legitimises relationships and connections with healthcare professionals, and the concept of palliative care, provides the context to trigger the mechanisms that produce the outcomes desired by children and families. At present, paediatric palliative care tends to run parallel to existing healthcare systems, with a lack of integration into other services (285), a situation that must be improved if all children with life-limiting and life-threatening conditions are to experience palliative care.

Determining when palliative care could provide benefit from this research was more challenging. Both the children and their family members had an awareness of the fragility of the child's situation, but this was often implied during discussions, rather than being spoken about openly. As described earlier in this chapter, the term "palliative care" was not viewed positively by the children and their families in this study. In previous research, it has been found to be associated with a specific service or phase in the child's illness, making it difficult to introduce as a new concept or service at a particular time for a child and family. There is wide debate about how overtly certain terms such as "palliative care", "end of life" and "hospice" should be used in conversation with patients (16, 18). The findings of an Australian study using conversation analysis have proposed that it may be possible to negotiate conversations about the end of life and dying with children and families without overtly referring to death (286). Previous research (a survey study) in oncology suggests that children and families may be more open to the idea of early referral to palliative care services than healthcare professionals believe they are (287). Children and families should have access to information about the palliative care that may be available to them from early on in the course of the child's condition,

including information about who will provide this. There were examples in this research of palliative care services being introduced as a routine part of care. One example was the introduction of palliative care as a routine process at the point of a relapse for a child with a malignancy. According to the clinicians who had implemented this initiative, it was working well. There are other examples in the published literature, including a study where a referral to a specialist paediatric palliative care team was made if a child was commenced on extracorporeal membrane oxygenation (ECMO) in intensive care (288). In Germany, the law states that every patient who has an incurable condition has the right to receive specialist palliative home care. Improved quality of care and higher levels of patient satisfaction amongst children and families have been reported since this legislation was introduced (102).

Identifying and negotiating the values of children with life-limiting conditions and their family members in relation to the term and concept of palliative care is a particularly emotionally challenging area of practice, requiring healthcare professionals to acknowledge the possibility that the child's condition is life-threatening and that they may die. This is further complicated by the "p word" problem, which is a pertinent issue in palliative care for children, since it is frequently conceptualised as a binary either / or option, despite broader definitions and policy advocating for a longer-term palliative care approach (289, 290). The values, acts and behaviours of healthcare professionals involved in providing care to children with life-limiting and life-threatening conditions and their families therefore has a direct impact on whether or not the children experience palliative care. More open recognition and discussion of the values of individual healthcare professionals and their capacity and ability to bear witness to the child and family situation at any particular time, would be helpful. The importance of the values of individuals in decision making in healthcare has been highlighted previously, with recommendations that decision making should combine knowledge from evidence-based medicine with the particular values of individuals (234). Values-based medicine (VBM) provides a useful framework and proposes that evidence-based healthcare would be implemented more effectively if the values of individuals were better identified and negotiated (291, 292).

Addressing the current inconsistencies in the conceptualisation, perception and understanding of palliative care for children may help the future delivery of this approach to all children who need it, including referrals to specialist paediatric palliative care services, where possible. Continuing to use broad definitions of palliative care requires a fundamental shift in culture and attitudes amongst all involved, if all children with life-limiting and life-threatening conditions are to experience a palliative approach to care (194). This depends on all healthcare professionals being able to face the possibility

that a child may die and having the capacity to deliver palliative care, a situation that this research proposes may not be a reality. It depends on the workforce who are motivated in this area of practice having access to adequate training, education and support in order to do so. Specialists in paediatric palliative care have an important role to play to legitimise the palliative care delivered by their non-specialist colleagues, role-model the approach, in the development of training and educational resources, and in ensuring that their specialist service offer is clear. Sense making of an intervention, and the development of shared understanding amongst stakeholders, are important steps in the implementation of a complex intervention, described in Normalisation Process Theory (NPT) (293), which explicates that in order for interventions to be effective, they must be capable of being widely implemented and normalised into routine practice. The components of NPT are coherence (sense-making of the intervention), cognitive participation ("buy-in"; commitment and engagement of individuals), collective action (the work that participants have to do to make the intervention function) and reflexive monitoring (reflection on and appraisal of the intervention) (293-295). As long as the "p word" problem persists, coherence, cognitive participation and collective action are unlikely, hindering the delivery of palliative care as both a broad approach, and the delivery of more practical aspects of care, such as a referral to palliative care services. Complexity science goes further to recognise the importance of self-organisation and adaptation amongst staff, and to encourage the sense-making process (263). This research provided evidence that engaged, committed senior leaders had a key role in securing organisational buy-in by placing value on palliative care, as well as securing financial resource in order to develop paediatric palliative care services. Senior leaders who recognise the complexity of palliative care, and provide staff with the opportunity to make sense, self-organise and buy-in to the delivery of palliative care for the children and families that they care for, could be highly influential.

Another option to address the "p word" problem would be to clearly assign the term "palliative care" to the specialist teams of the future, and focus less on the broad definitions. This is in keeping with the views of the children and family members who took part in the study, and seems to be the direction of some policy (19) and current research in the UK. In international healthcare systems, such as in the USA, the term "palliative care" may be more closely and clearly associated with specialist services. The further development of specialist paediatric palliative care services is a frequent focus of policy in children's palliative care, including NICE Guidelines in England (20, 196, 296). This is an important consideration as the numbers of children with life-limiting and life-threatening conditions rises and global estimates of serious health-related suffering, including in children, increase (297). Assigning the term clearly to specialist services, and placing less emphasis on broad definitions, may allow further clarification of the roles and responsibilities of the specialist

team. There is an increasing need for expertise, and research, in areas of practice such as complex symptom control and managing the end of life, including the withdrawal of life-sustaining treatments, areas of practice that raise difficult clinical and ethical issues (298-301), and may be the domain of specialists. One potential strategy through which all children with life-limiting and life-threatening conditions could receive palliative care is to fund sufficient specialist services and training to ensure adequate numbers of specialty-trained professionals. Currently the financial resource for the development of specialist paediatric palliative care services (defined as those supported by a consultant in paediatric palliative medicine) does not appear to be forthcoming, in the UK NHS or elsewhere. Regardless of the finances available, the development of the specialty as a distinct and separate entity, without careful consideration of the wider, complex and dynamic healthcare system, risks adding to the fragmentation of the system experienced by children and families, and may exacerbate the "p word" problem. The programme theory highlights how important it is to carefully consider the development of specialist services as part of the wider, changing, dynamic healthcare system. Future service design must recognise that the essential foundations of palliative care are well co-ordinated, holistic healthcare, where the preferences and interests of the children and family are at the centre. Responsibility for the provision of this holistic care should be taken at an organisational (meso and macro-system) level, and at a micro-system level, amongst all of the child's healthcare team, in hospital and in the community.

11.3. Methodological considerations

This section of the chapter describes the methodological considerations of the research. It begins with a description of my experience of the research, and the relevance of my subjectivity to the design and conduct of the research. The patient and public involvement (PPI) is then discussed, followed by the methodological strengths and limitations of the research.

My experience as a researcher
The roles, perspectives and motivation of a researcher influence qualitative research. In contrast to positivist, quantitative research, where objectivity is valued and bias affects whether or not the research findings are trustworthy, qualitative research positively values the subjectivity and reflexivity of the researcher. Reflexivity is an essential requirement for good qualitative research, with the researcher bringing their subjectivity to the process, recognising it and critically reflecting on both the knowledge that is produced, and the role that they have in producing that knowledge (219).

The introduction and methodology chapters (Chapters 1 and 4 respectively) of the thesis provided opportunity for me to outline the clinical experiences that have motivated this research, and my

epistemological position. The delivery of palliative care for children, who have complex healthcare conditions and family situations, in a complex and dynamic healthcare system, is a subject area that had caught my attention in my clinical practice as a GP and motivated my ambition to undertake high quality, policy-relevant research with the aim of improving care. Throughout the research, I have been aware of my subjectivity and the influence this has on the research process. I have also been aware of how the research procedures, particularly the interview and focus groups, may have influenced the research. I kept reflective notes diaries and had regular supervision throughout the research process, which have enhanced both my functional reflexivity (reflection on the research procedures), and my personal reflexivity (my personal experiences and circumstances, and how they influence the research).

My clinical experience as a GP was influential in both the design and conduct of the research. The fieldwork required time and flexibility, so the interviews were conducted only when it was convenient to children and family members. Although the interview participants knew that I was a healthcare professional, I was not directly involved in their clinical care. Their motivation to take part in the research was similar to mine, in that we shared an intent to improve services, and it often felt that we were working together. A similar dynamic enabled the focus group discussions. As a GP, and not a member of a paediatric palliative care team, I was viewed as a critical friend, with participants appearing able to speak openly about their experiences. Paediatric palliative care is an area of practice that GPs have little exposure to during training or in practice, so I had a lot to learn and consider through the course of the research, which helped me to maintain objectivity.

My GP training and professional development had equipped me with the advanced communication skills necessary for the interviews and focus groups, including rapport building, reflecting back, active listening and the use of open questions. I was in a position to be able to explore areas with children and families that were potentially very sensitive and difficult. It helped that I had a certain degree of "insider" knowledge of the healthcare system, being able to relate to some of the issues that were raised during both the interviews and the focus groups.

Meeting clinical teams to discuss the study and engage them in recruitment, and subsequently the data collection, took me into clinical environments, including hospital departments, and to the premises of community teams. These experiences provided an opportunity to develop insights into aspects of the experiences of children, families and healthcare professionals that were not specifically part of the data collection. These clinical environments were completely different from my usual clinical work setting, the GP surgery, and I often reflected on the different kind of "normal" that existed for professionals working there. When everyday work life involves caring for critically

unwell children, the perspective of a professional on whether or not a condition is considered life-limiting or life-threatening, seems very different to mine as a GP, who sees these children less frequently in clinical practice. For specialist paediatricians and paediatric teams, providing care for children with life-limiting and life-threatening conditions is an everyday norm, and this perspective is likely to influence clinical decision making in relation to palliative care.

The data analysis, particularly the analysis of the child and family interviews, required a level of personal emotional awareness and resilience that I had underestimated. The serial interviews allowed me to develop rapport with the children and families, and reading back their transcripts, taking time to reflect on what they said, was more emotionally challenging than I had anticipated. During this time, as well as an ongoing process of reflection, it was necessary to seek extra psychological support from university counselling services. I was surprised to find myself learning about and reflecting on my own priorities, including the experiences of my own (well) children, and the choices we were making as a family, as I undertook the data analysis.

A further important area for reflexivity during the research was my interaction with policy makers at a local, regional and national level. Alongside my clinical practice and research, I have taken on a variety of different policy roles. Attendance at meetings and becoming progressively more involved in the development of local, regional and national policy required a further process of ongoing reflection, through diaries and supervision. My overall experience has been that policy makers welcome insights from research and the potential for new ideas, however there have been many occasions where the complexity and continually changing nature of the healthcare system and influences within it have felt overwhelming. The ability to reflect on these concerns throughout the research process has been valuable, informing my desire to ensure that my research, now and in the future, focusses on the experiences of those receiving care, and is policy-relevant and applicable in a complex healthcare system.

Patient and Public Involvement

A significant strength of the study was the Patient and Public Involvement (PPI) that was integral to every stage of the research, from the research design stage through to dissemination. The PPI incorporated the views of children and young people from a variety of backgrounds, and with a diverse range of interests, experiences and expertise.

PPI led to a change in the study title so that it did not include the term "palliative care". This provided opportunity to place an emphasis on the wider experiences of healthcare of children with life-limiting or life-threatening conditions (described in the participant information sheet as those "that may or may not get better"), and their families. Broadening the scope of the research in this

way allowed for in-depth consideration of how palliative care is conceptualised and delivered within the wider healthcare system, and why some children receive care for specialist paediatric palliative care services, while others do not.

The PPI also led to further research activities that took place alongside the PhD study, including a survey project about the language of palliative care, and a variety of dissemination activities. These have included the young people leading oral presentations and presenting posters at national and international conferences (detailed in the PPI timeline in Chapter 2 and in Appendix 7). PPI group members advised on the development of presentations, including a comic strip presentation (Appendix 8) and the research recommendations. There are plans to develop a short dissemination film in partnership with PPI group members.

Methodological strengths and limitations of the empirical research
The research carried out for this study following the literature reviews was small in terms of participant numbers, but through serial interviews provided highly detailed, in-depth insights and increased understanding into the experiences of healthcare, and palliative care, of children with life-limiting and life-threatening conditions. It has captured the views of the children and has used realist methods to describe the interactions and relationships that are critical to the delivery of palliative care within a complex healthcare system. This section provides a discussion of the strengths and limitations of each stage of the empirical research. The strengths and limitations of the literature reviews have been discussed in Chapter 3.

Study population
The inclusion criteria of the study did not dictate that children and families had to be under the care of a specialist paediatric palliative care team in order to take part. This was a strength of the study in that it provided opportunity to explore the facilitators and barriers to discussions about palliative care and referrals to specialist services during the interviews. It did, however, raise some ethical concerns, and required careful navigation during the interviews. As a researcher, I did not raise the term or concept of "palliative care" with families unless there was a cue within the interview to do so. As outlined in the findings of the study, the life-limiting or life-threatening nature of a child's condition was often implied rather than being openly addressed during the interviews.

A significant strength of the study is that children were included in the interviews, which addresses an important gap in current research. Ten children participated in interviews. This study population is relatively small, and the dataset is therefore relatively limited. This study, unlike others, did include the views and perceptions of children, and conducting serial interviews resulted in the collection of in-depth insights into their views, perceptions and experiences. Furthermore, there

were strengths in the diversity of the study population in terms of age, ethnicity and the range of different conditions with which the children were living. Four of the children were unable to participate, as they were unable to communicate verbally. Their family members were interviewed, which was considered appropriate since their views regarding healthcare and palliative care are important. Research to investigate the views of children who cannot communicate verbally is an important area for future research, and there are researchers who are conducting work into how to involve children who can't communicate verbally in research. The results of their work will have implications for the design of future research in children's palliative care and other aspects of children's health and social care delivery (302, 303).

A further potential limitation is that the sample is likely to have represented the children and families who were most motivated to participate, and were all recruited via a children's hospital or children's community team in a single city in the UK, and may therefore not be representative of a wider population. Other potential limitations of the study included the exclusion of neonates, pre-school children and young people over the age of 18 years who are making the transition to adult services. These groups all warrant research in their own right. Given the time and resource constraints of the study, a decision was made to conduct all of the interviews in English, which presented a further potential limitation. English was the first or second language for the children and families who took part. Further research into the experiences of children and families who cannot communicate in English is necessary. There is also an ongoing need to investigate the views of fathers. More mothers than fathers took part in the interviews. Fathers were given the option to take part but were often unable to do so due to work commitments, which is an important consideration for the design of future research. Two brothers took part in the interviews. Their contributions were included in the data analysis, as the views of family members, but this data is too limited to draw any firm conclusions specifically about the views of brothers or siblings.

As in previous research with children and families (111, 304) and in palliative care research (247), gatekeeping was a relevant factor in recruitment, with some specialist teams taking weeks or months to decide whether to ask families about the project. The intention of the recruitment strategy was to allow children and families to make the decision about whether they wanted to take part or not, rather than for staff to make this decision for them. Certain teams within the hospital and community were more proactive in providing children and families with the study information. Half of the families were recruited via two teams, the community children's palliative care team and the hospital acute pain team. The reasons why these teams were more engaged are unknown but may have been related to their perceived need for the research. At the time of recruitment, the

hospital palliative care team was in development, and the acute pain team, for example, were involved in the care of many children with complex life-limiting and life-threatening conditions.

Some patient groups were notable by their absence from the study population, including children with liver disease and cardiac disease. This was despite presenting the research at their departmental research meetings and having one-to-one discussions with nursing staff. Other groups who were notable by their absence were children and young people with conditions such as Duchenne muscular dystrophy and cystic fibrosis, both of which are specific examples of life-limiting condition in category 2 of the Together for Short Lives categories. These conditions are now associated with much longer life expectancies than they were when the categorisation was proposed, with many young people affected by these conditions making the transition to adult services (305, 306).

A pragmatic approach to recruitment for the focus groups was taken, with the focus group discussions being conducted at existing children's palliative care network meetings. This was a successful approach, recognising that busy healthcare professionals may not always be able to take part in extra research activities. The groups varied in size and diversity, with participants from a range of professions present. Balancing the dynamics of each group so that all who wished to could have a say required careful negotiation, active listening and watching for non-verbal cues from participants. It is possible that within such large groups, there were participants who did not feel able to participate. The setting of a children's palliative care network meeting may also have influenced the contributions that participants made, if they felt that they should respond in a particular way in the presence of their network colleagues. This was not my perception of what was happening, as many participants shared a wide range of views and reflections. Written feedback sheets were provided in an attempt to mitigate against this possibility and to capture any other ideas and thoughts that participants wished to raise.

A range of healthcare professionals took part in the focus groups. The majority were doctors and nurses, and a small number were allied healthcare professionals, including play therapists, pharmacists and clinical psychologists. The contributions of all have been included in the analysis, as they are all healthcare professionals involved in palliative care. Furthermore, data from the interviews with children suggested that allied healthcare professionals and other hospital staff, including catering staff and domestic staff, play a key role in the care that they receive. With a smaller number of allied healthcare professionals taking part in the focus groups than other professional groups, it should be recognised that data relating specifically to their perspective is limited and does not necessarily represent the views of allied healthcare professionals more widely.

Data Collection

A methodological strength of the study was the in-depth, contextual, qualitative nature of the data, with multiple child and family member stories captured over time, through serial interviews. A strength of this study is that it followed families through different stages of illness. For some, the life-threatening condition became more stable through the course of the study, or went into remission. For others, the child died. As anticipated, the child and family stories were highly individual, and it is likely that further new insights would be obtained, without reaching data saturation (the point where no new insights are provided through further interviews) around the themes, had the interviews continued (307).

The healthcare professional focus groups only included professionals who worked in some way in palliative care for children, or who had an interest in this area of clinical practice. Gathering the views of the wider workforce, including those who are less engaged with children's palliative care would be valuable. A small number of system leaders in management roles took part in the focus groups, but there is more to be understood about the perceptions of policy-makers and senior managers in healthcare about palliative care.

Data Analysis

Thematic analysis was appropriate for analysis of the in-depth data gathered from the serial interviews. One of the strengths of the analysis is that the themes were developed iteratively from the data, keeping a focus on the subjective experiences of the participants. The application of a realist logic to the thematic analysis is a novel approach to research in palliative care for children, but is appropriate as it recognises the complexity of palliative care as an intervention. A particular challenge with a realist approach in this study was defining "palliative care". Much previous realist research has evaluated more discrete interventions. During the analysis, the research team regularly reflected upon and debated definitions of "palliative care" and how it differed (or not) from holistic, person-centred healthcare. During the data analysis, we found that palliative care was most often conceptualised as a specialist service by participants, rather than as a broad approach or philosophy of care. Ensuring clarity and consistency in how the term "palliative care" has been used through this research has presented a challenge, requiring regular reflection and consideration with the supervisory team.

One of the strengths of the realist approach is that it provided a practical method through which to describe some of the unseen complex and nuanced elements of healthcare and palliative care delivery to children. The dataset from the empirical research, although detailed, is limited in that the number of participants was relatively small, however it has been possible to use this data refine and

refute the findings and the programme theory proposed as a result of the realist literature review. The power of realist research lies not necessarily in the size of datasets, but in its explanatory nature, going beyond what can be measured in the social world, to explain the deeper, causal powers that shape what can be observed (211). Through the development of CMOCs, and description of mechanisms, it has been possible to provide insights into the outcomes desired by children and their families, and to devise explanations about how these can be achieved in certain contexts. Best practice guidelines in realist research have been followed, and all of the CMOCs presented through the research have been configured, with each context linked with an outcome via a mechanism to produce an explanatory configuration. It is a requirement of realist research that CMOCs are precise and narrow and can therefore be used to develop implementable or testable recommendations (258). The CMOCs and the overarching programme theory, which together outline the micro, meso and macro-system factors necessary in order for the complex intervention of palliative care to children to be delivered, is testable. The definition of a "mechanism" is an area of debate amongst realist researchers. Mechanisms can be difficult to describe because they are hidden. For the purposes of this research, a mechanism was defined as a description of how specific outcomes of a programme are achieved in certain contexts (126).

A further strength of the realist analysis was that the development of the overall programme theory drew upon multiple sources of data (literature reviews, serial interviews and focus groups). Bringing together these multiple sources of data to provide detailed contextual accounts with contrasting and complementary viewpoints was a method to bring rigour to the analysis. The generalisability of this realist research is enhanced by drawing upon existing theory, including complexity theory, to support the claims made.

Both the thematic analysis and the realist analysis would be stronger with more sources of data. Many more potential CMOCs could be constructed if the research had included more children, children with a more diverse range of conditions, members of the extended family, and healthcare professionals who do not have an interest in palliative care. It is important to acknowledge that a programme theory is only ever partial. However, through taking the realist approach and devising a programme theory, this study has addressed the research questions and aims to make a clear contribution to policy development.

There are quality standards for the conduct of realist research, the RAMESES standards. A table detailing how this study meets those standards is provided in table 11.1 below:

Table 11.1: Application of RAMESES quality standards to the research (table adapted from RAMESES standards and Papoutsi et al (128, 129):

Quality criteria	How the criteria were fulfilled
The research purpose	
The research topic is appropriate for a realist approach	Palliative care is a complex intervention that requires the active input of individuals, whose role is influenced by other individuals including patients and colleagues. Palliative care services are embedded in other social infrastructures (such as hospitals, hospices and primary care) and affected by institutional and system factors (such as local and national policy guidance and commissioning). One of the aims of the research was to produce policy relevant recommendations, which is one of the specific aims of realist review.
The research questions are framed to be suitable for realist research	The research questions broadly ask "when" and "how" palliative care provides benefit to children and their families. This was refined further to specifically ask about the mechanisms by which palliative care provides benefit, and the contexts in which these mechanisms are triggered, providing an explanation for when, how and why palliative care is beneficial.
Understanding an applying a realist principle of generative causation	
A realist principle of generative causation is applied: The research demonstrates understanding and application of a realist philosophy and realist logic that underpins the analysis	The research follows the RAMESES standards for realist evaluation. A realist logic of analysis allowed for contexts, mechanisms and outcomes to be identified in the data, with a focus on generative causation and the subsequent development of CMOCs. This research applied realist logic to a broad and indistinct area of practice rather than a discrete intervention.
Constructing and refining a realist programme theory or theories	
An initial tentative programme theory (or theories) is identified and developed. The programme theory is "re-cast" as realist programme theory.	The initial programme theory (PT) was derived from policy documents and a systematic review. A more refined PT was developed through realist review of the literature. This has been refined and refuted further in order to produce the PT and policy relevant recommendations. There has been a continual process of engagement with stakeholders and PPI throughout the research.
Research design	
The research design is described and justified.	The research design has been explained and justified throughout, with a clear rationale provided. Protocols have been published in PROSPERO for both literature reviews, and a protocol for the qualitative study published in BMJOpen.
Ethical clearance is obtained.	Ethical approval was granted by the UK Health Research Authority on 14th September 2016 (IRAS ID: 196816, REC reference: 16/WM/0272, Sponsor: University of Warwick).

Data collection methods	
Data collection methods are suitable for capturing the data needed	The data collection methods (serial interviews and focus groups) were designed in close collaboration with a PPI group. The interviews were designed to be open and reflexive to the needs and changing situations of children and families. Focus groups were also semi-structured, allowing for iterative development of the topic guide both during and between each focus group, in order to capture key themes.
Sample recruitment strategy	
The respondents or key informants recruited are able to provide sufficient data needed for the realist research.	The children and family members who took part in the study all had relevant experiences of life with a life-limiting or life-threatening condition, and healthcare services. The focus groups were designed to capture a diverse range of healthcare professionals views at a time that was convenient for those taking part.
Data analysis	
The overall approach to analysis is retroductive	Mechanisms have been abstracted from the findings of research.
Data analyses processes applied to gathered data are consistent with a realist principle of generative causation	The focus of the realist analysis has been explanation and generative causation.
A realist logic of analysis is applied to develop and refine theory	The steps taken in the realist analysis have been explained and are in keeping with the RAMESESII guidance.
Reporting	
The research is reported using the items listed in the RAMESESII reporting standards	The RAMESESII standards will be used to structure academic papers of the research.
Findings and implications are clear and reported in formats that are consistent with realist assumptions	The findings have been presented using diagrams to assist with clarity, and close attention has been paid to ensuring that the recommendations and new model are clear and understandable. PPI has informed the reporting of the research findings.

11.4.   Implications for policy and practice

This research has been conducted from an epistemological perspective of realism, acknowledging that both the material and the social worlds are real, independent of, and inter-dependent with, human understanding (210). The realist approach goes beyond what can be measured in the social world, to explain the deeper, causal powers that shape what can be observed, and places emphasis on understanding the explanations for *how* and *why* things work (210, 211). Realism also recognises that social systems, such as healthcare systems, are complex, dynamic and open systems that cannot be controlled. For the children who took part in this study and their families, the life-limiting and life-threatening conditions with which they lived, and the impact that this had on their family situations, were complex and unpredictable. The healthcare system, a complex system made up of people and processes, also constantly evolved and changed around them (308). Complex systems evolve through self-organisation (actions of individuals at the frontline in response to culture and available resource), interactions between interdependent parts of the system, and sense-making (the process by which people, as individuals and groups, assign meaning to experience and link it to action) (263).

There is a growing evidence base to suggest that palliative care can improve the quality of life of children with life-limiting and life-threatening conditions, however such care is provided inconsistently, and the availability of specialist services is patchy in the UK and internationally. The aim of this research was to provide new insights and understanding into the healthcare experiences of children with life-limiting and life-threatening conditions, and their families, in order to understand how palliative care can be delivered more consistently to children who could benefit from it. The research has limitations, but contributes to the evidence base to inform future policy and practice in palliative care for children. The realist approach tested the initial programme theory that palliative care "works" through literature review and empirical research. New theories, including a new programme theory, have been generated. Complexity science underpins and informs the research and is highly relevant to the discussion of the implications of the research.

The programme theory proposes that whole system change is healthcare is required in order to improve the delivery of palliative care to children, taking into account the complex situations of children and their families, a need to make palliative care possible, and an ambition to develop specialist paediatric palliative care services, considered in more detail below:

The complex situations of children and their families
The children who took part in this research were living with individual, complex medical conditions, associated with a high level of unpredictability. Their clinical care and management often involved new medical treatments and technologies. Their conditions did not fit neatly into the current

categorisation of palliative care need (2). Furthermore, there is complexity provided by the individual family situation and the impact that the child's condition has on family life. Family situations, and the implications of the child's condition on family life, were also highly individual factors that added to the complexity and unpredictability of the child's situation. Furthermore, communication with children and families about the life-limiting or life-threatening nature of the child's condition, including palliative care, was complex.

Children and families sought to maintain as normal a life as possible in their abnormal circumstances. They described a variety of sources of peer support when their child became unwell. Some of the family members were involved in providing support and education for other families. Others sought support from the families of children with similar conditions on the wards, and on social media. It is important to recognise the diversity and influence of these sources of support, how family members access support, and the impact that different sources of information can have on their expectations and hopes. More work is required to understand the opportunities and threats associated with wider societal influences and sources of information. These have been pertinent issues in high profile cases (54), and are likely to add further to the complexity of ethical, clinical and emotional dilemmas in the management of children with life-limiting and life-threatening conditions and their families in the future.

Making palliative care possible

Palliative care is a complex intervention that needs to be delivered in the context of the increasingly complex needs of children and their families, and an emergent, dynamic healthcare system. The proposed programme theory outlines a whole system approach, with important factors at the micro, meso and macro-system levels all needing to be in place for children and their families to experience the outcomes most important to them.

The provision of care to children with life-limiting and life-threatening conditions, who may die, is an emotionally and ethically demanding area of practice. This research has described a combination of healthcare professional contexts that need to be present at the micro-system level in order for healthcare professionals to be able to deliver this care, whether or not it is formally labelled "palliative care".

Professional contexts included personal motivation, an innate ability to bear witness to the child and family situation, and an ability to form connections with children and family members. Listening and advocacy were important interventions in their own right. It follows that those who have the ability and personal capacity to recognise the possibility that a child may die, and who are able to be

alongside families and bear witness to their situation, are more likely to do so if their working environment allows. The children and families gave multiple examples of instances when this appeared not to be the case, due to a fragmented and rigid healthcare system, where it appeared more culturally acceptable to continue to propose medical treatments or onward referrals to other specialists than to face the prospect of the death of a child. Continuity of care was an important factor contributing to the development of relationships between children, family members and healthcare professionals. Sometimes this was relational continuity with a particular healthcare professional. At other times, this was through consistency of healthcare professionals in a particular healthcare environment. Both are important factors for consideration in the design of healthcare services.

The research highlighted the emotional impact that the provision of palliative care to children can have on healthcare professionals. Careful consideration should be given to nurturing and supporting healthcare professionals with the emotional impact of the provision of such care. A fundamental shift in attitudes and culture in healthcare, to allow more open dialogue about the support and care needs of staff, whether they can bear the prospect of a child dying and provide support to families, is needed. There may also need to encourage more open discussion at times when healthcare professionals feel they do not have the personal capacity to bear witness to the child and family situation.

Healthcare professionals who took part in the focus groups highlighted training and education as a tangible strategy to improve palliative care. While education can have a positive impact on palliative care knowledge and attitudes (309), the values, motivation and previous experiences of individual clinicians also has an impact on their professional behaviour (234).

Complexity science outlines the concept of self-organisation at a micro-system level as an important step to the implementation of a programme or intervention. Self-organisation is a process by which relationships with a concept, intervention or programme may form, in changing systems such as the healthcare system, created by feedback mechanisms that can be positive or negative. This process is inevitable according to the structures and resource at a micro-system level. In palliative care for children, this would include recognition of how clinicians within teams at the micro-system level respond (self-organise) within a healthcare system that is rigid and fragmented, and within a death-defying, death-denying organisational and societal culture. Clinical experiences of caring for children with life-limiting and life-threatening conditions, and their families, and working with specialist paediatric palliative care teams, is likely to have an impact on healthcare professionals, which could be positive or negative depending on the situation, the individuals involved, and the outcome. These

experiences, and the resulting self-organisation within the micro-system, may affect whether or not professionals feel motivated to deliver palliative care in the future.

Role-modelling was highlighted by healthcare professionals as an important and significant responsibility in challenging the "blind spot" of palliative care amongst their colleagues. The development of palliative care "champions" in adult healthcare settings has been found to have beneficial effects including increased awareness of palliative care need, more communication with patients (310), and increased levels of confidence in palliative care amongst staff (311). In children's palliative care, there may be opportunity to develop a similar role amongst professionals who are particularly motivated and committed to the provision of palliative care, who could take a lead on the provision of palliative care within their specialty. These individuals could be supported by specialist paediatric palliative care services, for education and peer support, and in turn provide peer support and education and act as role models for other professionals within their speciality. Their presence would have the potential to change culture within their speciality; a network of champions could potentially contribute to culture change across an organisation or community.

Language in palliative care is an area of considerable debate (16, 17). The children, families and healthcare professionals who took part in this research all described a "p word" problem. Complexity science describes sense making as an important step in the implementation of a programme or intervention (312). In the case of palliative care for children, sense-making of the situation should allow stakeholders to express their concerns and understanding or lack of knowledge related to the term "palliative care" and what it might mean for a child and family. Children and families could also be encouraged to express their preferences around the language used in palliative care more openly. Other principles of complexity science in healthcare are useful to consider in relation to language and palliative care, including adaptive capability in staff, and ensuring that conflicting views are viewed as an opportunity to develop new solutions (263). Internationally, there are several examples of services that have moved away from the term palliative care to other names for their services, including CompassionNet (97), and the Paediatric Advanced Care Team (PACT) (313).

Increasing the provision of specialist paediatric palliative care services
The development of specialist paediatric palliative care services within an organisation requires additional financial resource, including funding to support the training and development of the medical, nursing and allied healthcare professional workforce. The evidence base to support the development of specialist paediatric palliative care services is growing, with some studies showing potential economic benefit (106, 107, 313), as well as improved care for children and families. There

is evidence from a randomised controlled trial in adult palliative care that the provision of specialist palliative care alongside disease-directed treatment can add to the life expectancy of patients (213). This makes an appealing case for palliative care for children. Learning from research of this nature should help to inform the language used in designing services, challenge the "collusion of immortality" and the perception that palliative care is not an "inspirational" area of practice.

Current models of service delivery, which have specialist services at the top of a pyramid (as in figure 1.1, Chapter 1), may risk adding to the fragmentation of the healthcare system. It is important that plans to develop specialist services are made within the wider healthcare system context, carefully considering the complexity of the wider system and in particular, key interdependent services in hospital and community care. The research findings have highlighted the fragmentation of the healthcare system that children and families can experience, with specialist paediatric palliative care services becoming part of that fragmentation, suggesting that the integration of specialist paediatric palliative care services into other services requires further attention. Specialist paediatric palliative care services have been proposed in the programme theory as one element of the whole system change needed in order to improve palliative care for children. The research provides evidence that their presence within an organisation leads to outcomes beyond service delivery, with role-modelling and legitimising palliative care as an approach contributing to the self-organisation and sense-making processes amongst individuals and teams at a micro-system level.

There was evidence in this research that the commitment and engagement of senior leaders, both clinicians and managers, who role model and legitimise palliative care, could be key to the development of new services and an organisational culture where palliative care is acceptable. The need for this leadership in palliative care is specifically described in national palliative care policy in England (49). Organisational leaders who display understanding of the value, benefits and importance of palliative care are required to work towards providing an organisational culture where palliative care is deemed normal and necessary. The commitment of senior leaders may also be necessary to ensure effective collaboration across healthcare settings, including with voluntary sector organisations, to make innovative use of the finite resources available to develop specialist paediatric palliative care services.

11.5.   Recommendations from the research

One of the key aims of this research was to produce policy-relevant recommendations. The recommendations are outlined below.

1. Whole system change is required in order to improve the delivery of palliative care to children

The programme theory derived from the findings of this research proposes that whole system change across the micro, meso and macro-system in healthcare services is required in order for children with life-limiting and life-threatening conditions, and their families, to experience palliative care. Placing emphasis on the ensuring that the contexts in which hidden mechanisms are triggered in order to produce important child and family outcomes are in place across the system could lead to improved experiences of palliative care.

2. Palliative care must be personalised to meet the unique, individual needs of children

Future policy and guidance in palliative care for children should place more emphasis on highly individual, unique child and family situations. The complexity and uncertainty that pervades for children, families and their healthcare professionals requires clear acknowledgement in policy and organisational procedures, with a dynamic and flexible approach allowing the adaptation of services to the individual needs of the child and family at any particular time. Current categorisation of palliative care need could be revised to incorporate the complexity and unpredictability associated with children's conditions and new medical treatments and technology.

3. Future policy in palliative care should place emphasis on the need for trusted relationships

Whether palliative care is defined as a broad approach or as a specialist service, the importance of established, trusted relationships between children, families and individual healthcare professionals should not be underestimated as the foundations for palliative care. The provision of a working environment where these relationships can be established, and where connections between children, family members and healthcare professionals that provide a feeling of security should be a priority for those involved in service design and policy. Future service design in children's palliative care should nurture and support professionals who are able to provide such care.

4. The specialty of paediatric palliative care should be integrated into existing services.

Future policy and service design should take steps to ensure that the development of specialist paediatric palliative care services includes integration into the wider healthcare system to avoid fragmentation. There is a need to maximise the opportunities for role-modelling and legitimising palliative care, as well as the development of skills and expertise to manage the increasing complexity of the conditions with which children live.

5. Unwavering leadership from healthcare system leaders is required for the development of palliative care for children

Every healthcare organisation should have a member of the senior leadership team, or board member, to oversee and take responsibility for palliative care within the organisation. Commissioners should hold healthcare organisations to account for having these members of the senior leadership team.

11.6.   Dissemination plans

The main target audience for this research is people who have decision-making capacity in healthcare. The research has been designed with the intention of providing recommendations that are of immediate relevance to policy makers and commissioners, with the intention of contributing to the evidence base to improve the delivery of healthcare to children with life-limiting and life-threatening conditions.

The dissemination and opportunities for impact that have occurred alongside the conduct of the research are summarised in Appendix 7. These include the publication of academic papers, presentations at national and international conferences with members of the PPI group, and opportunities to contribute to policy development at regional and national levels. Academic outputs have included four peer-reviewed papers from the thesis so far (Appendix 1), one of which has been cited in national policy guidance (19). Several more are planned, including papers of the interview and focus group findings, a methodological paper about how the realist approach was used, a PPI paper, and a paper outlining the recruitment strategy and challenges. A film to accompany the dissemination of the research findings is planned with the PPI group.

11.7   Suggestions for future research

There is a paucity of research in paediatric palliative care to support the future development of services (314). New research questions have arisen throughout the course of this PhD, and some research to address these is already underway. This research and further suggestions for future research are outlined below at micro, meso and macro-system levels.

1. Research to investigate micro-system factors

Language, and the use of the term "palliative care", has been a pertinent issue throughout this research and is an area for further consideration. Research to investigate the views and perceptions of children and young people of the term "palliative care" including a survey of children, young people and healthcare professionals in five UK children's hospitals, has been carried out alongside this PhD research, and preparation of a paper for dissemination of these findings is underway.

This study has included children, and provided new insights into their views and perceptions. Gatekeeping by healthcare professionals was a relevant factor in recruitment of children to this research. It would be helpful to conduct research to understand more about how and why healthcare professionals will, or will not, assist with recruitment of children with life-limiting and life-threatening conditions to research. These factors may include workload, culture, and attitudes towards research and palliative care within a specialty. Furthermore, there is more work to do to ensure that children and their families have access to information about research so that they can make autonomous decisions about whether or not to take part.

Much more research that includes children with life-limiting and life-threatening conditions as participants is required, including with children who make use of non-verbal communication methods. There is also a need for further investigation into the delivery of palliative care for children in culturally diverse populations and those in complex social situations including looked after children, those for whom children's safeguarding is a concern, and children and families living with poverty and homelessness. Most of the family participants in this research were mothers. Research to investigate the views of fathers, and extended family members would also be of value.

Further investigation into the impact of children's condition on all aspects of their life would be valuable. This includes research to understand the child's experience of symptoms, and living with complexity and unpredictability. The need for further research into specific aspects of palliative care, including symptom control, optimal medication regimes and methods of administration have all been highlighted previously (314). The evidence base to understand and inform communication between healthcare professionals and children and young people about their diagnoses, priorities for life and advance care planning is limited and more research is required to understand this complex area. This should include the navigation of personal preferences for communication, how and when information can be delivered to and shared with children at different developmental stages and with different diagnoses, and the impact of relationships with healthcare professionals on that communication. Other complicated aspects of communication with families, such as collusion and the unintended consequences of following parental preferences for the information that is shared, or not, with their children would also be of value.

Research to map the unpredictable, multi-dimensional impact of life-limiting and life-threatening conditions on children's lives, including the impact on their psychological development and education, and specific cultural and religious needs of children and families, would provide exciting new insights to inform the delivery of responsive healthcare services, including palliative care, in the future. Alongside this, more research to understand the experiences, perceptions, benefits and risks

of interventions such as advance care planning, is an area for future research, particularly given the emphasis placed on such interventions in current policy and guidelines (196).

An unanticipated finding of this research was the use of online communities and social media by children and families to communicate their situation and to seek information and peer support. This is an important area for future research, including consideration of online platforms for data collection. Research to understand social media as a source of information and peer support, and how online discussions can affect a child or family members understanding and expectations of their experience is an emerging issue. The response of healthcare professionals to the scenarios described on social media, and whether or not they have a professional responsibility to ensure a healthcare response to certain situations requires careful consideration and research in the future.

There is also research to be done to further understand the experiences of healthcare professionals delivering palliative care to children, both specialists and members of the wider workforce delivering a palliative approach to care. This research has highlighted the need for established, trusted relationships with healthcare professionals. Communication, and the consultation, between children, families and healthcare professionals is an important area for further investigation, particularly in relation to navigating the "collusion of immortality" within a fragmented healthcare system.

Given the highly emotional aspects of providing care to children with life-limiting and life-threatening conditions, and current workforce concerns, research to examine how best to nurture, support and care for the workforce would be highly valuable. Children's palliative care provides an interesting and complex area for research to examine psychological support for staff, team working and the provision of continuity of care, and the impact that this has on factors such as patient experience, patient safety and the experiences of healthcare professionals.

2. *Research to inform healthcare service development at the meso-system level in palliative care for children*

Specialist paediatric palliative care is a relatively new specialty, and there is a need to continue to grow the evidence base to support its development. Research in adult patients with incurable lung cancer has shown an association between the provision of specialist palliative care and an increased life expectancy (213). Any similar association between the provision of specialist paediatric palliative care services and longer life expectancy in children would provide a powerful narrative in the provision of palliative care for children. Economic analysis of specialist paediatric palliative care services is required in order to understand any potential benefit to the healthcare system.

There is a need to conduct research into palliative care delivery within the wider healthcare system in order to understand and inform the relationships between specialist paediatric palliative care and other services, and their potential for effective integration given the "collusion of immortality" and the fragmentation of the healthcare system that exists. This includes interactions with other services in hospitals and the community, both in health and social care. A subanalysis of the qualitative data from this study related to child and family experiences of primary care in palliative care for children is underway. This research did not explore the family experiences of antenatal or neonatal palliative care, or young adults who are making the transition from paediatric to adult services, which are both important areas for future work.

3. *Research into macro-system factors in the provision of palliative care for children*

The programme theory generated as a result of this research proposes factors at a macro-system level as an important part of the whole system change required to improve palliative care for children. Further research to understand the views of senior leaders in healthcare, policy makers and senior managers of children's palliative care would be of interest, including research to understand more thoroughly the factors that may currently constrain or facilitate the development of services, beyond financial resource. This is an area that is rarely explored in research, but warrants further attention.

11.8   Chapter summary

Chapter 11 has provided a discussion of the research findings, situating them within the current literature to consider the contribution of this research to knowledge. The research findings and programme theory have been used to produce policy-relevant recommendations. This leads on to the conclusion of the thesis in Chapter 12.

12.   Conclusion

Palliative care is a complex intervention that can improve the quality of life of children with life-limiting and life-threatening conditions and their families. This is an important area for consideration in international healthcare systems, since the number of children with such conditions, many of which are complex and highly unpredictable, is rising.

The focus of the research has been the child and family experience of healthcare when a child has a life-limiting or life-threatening condition. The research has examined the current evidence base for policy and practice, and built upon that by gathering rich accounts of the experiences of children, their family members and healthcare professionals. It has not just described the problems within current healthcare for children with life-limiting and life-threatening conditions and their families, but through a realist approach, has led to the description of the many contextual factors that must be in place in order to trigger the hidden mechanisms that lead to desired outcomes for children and their families. Bringing together the literature reviews and the findings of the empirical research has allowed for the generation of a new programme theory. This theory has formed the basis of policy relevant recommendations that are intended to assist the translation of palliative care policy and guidelines into practice.

Important contexts exist at micro, meso and macro-system levels in healthcare. Children wish to lead as normal a life as possible in their abnormal circumstances. Each child is unique; they have individual needs and family circumstances. Family members take on the role of carers, and are experts in the care of the child, but are vulnerable given the complexity and uncertainty associated with the children's conditions. At a micro-system level, established, trusted relationships between children, family members and healthcare professionals who have the ability and capacity to bear witness to their situation are fundamentally important. This may be better achieved within a work environment that places emphasis on connections with healthcare professionals, consistency and continuity of care, and with the support of specialist paediatric palliative care professionals. These important considerations for future service design will depend upon system leaders recognising the complexity of this clinical practice area. Improving palliative care for children requires a whole system approach to service design and commissioning, with greater emphasis on the need for trusted relationships. Training, education and support should be available to nurture professionals who have the motivation and capacity to provide palliative care, and specialist paediatric palliative care should be integrated effectively into existing services. The implementation of new models of palliative care must be accompanied by robust research and evaluation.

References

1. World Health Organisation. http://www.who.int/cancer/palliative/definition/en/
2. Together for Short Lives. Children's Palliative Care Definitions. https://www.togetherforshortlives.org.uk/changing-lives/supporting-care-professionals/introduction-childrens-palliative-care/categories-of-life-limiting-conditions/.
3. Mitchell S, Slowther A-M, Coad J, et al. Ethics and Patient and Public Involvement with Children and Young People. Arch Dis Child Ed Pract, Published Online First: 09 February 2018. doi: 10.1136/archdischild-2017-313480
4. Mitchell S, Morris A, Bennett K, et al. "Specialist paediatric palliative care services: what are the benefits?" Arch Dis Child Published Online First: 04 April 2017. doi: 10.1136/archdischild-2016-312026
5. Mitchell S, Bennett K, Morris A, et al. Achieving beneficial outcomes for children with life-limiting and life-threatening conditions receiving palliative care and their families: A realist review. Palliat Med https://doi.org/10.1177/0269216319870647.
6. Mitchell S, Slowther A-M, Coad J, et al.. The journey through care: study protocol for a longitudinal qualitative interview study to investigate the healthcare experiences and preferences of children and young people with life-limiting and life-threatening conditions and their families in the West Midlands, UK. BMJ Open. 2018;8(1).
7. Fraser LK, Miller M, Hain R, et al. Rising national prevalence of life-limiting conditions in children in England. Pediatrics. 2012;129(4):e923-9.
8. Cohen E, Kuo DZ, Agrawal R, et al. Children With Medical Complexity: An Emerging Population for Clinical and Research Initiatives. Pediatrics. 2011;127(3):529-38.
9. Langton-Gilks S. Follow the Child. Planning and having the best end of life care for your child. London & Philadelphia: Jessica Kingsley Publishers; 2018.
10. Aldridge J. Living with a Seriously Ill Child London: Sheldon Press; 2007.
11. Fraser L, Parslow R, McKinney P, et al. Life-limiting and life-threatening conditions in children and young people in the United Kingdom. Pediatrics. 2012;129(4):1-7.
12. Beecham E, Hudson B, Oostendorp L, et al. A call for increased paediatric palliative care research: Identifying barriers. Palliat Med. 2016;30(10):979-80.
13. Hain R, Heckford E, McCulloch R. Paediatric palliative medicine in the UK: past, present, future. Arch Dis Child. 2012;97(4):381-4.
14. Murray S, Kendall M, Boyd K, et al. Illness trajectories and palliative care. Br Med J. 2005;330:1007-11.
15. Hawley PH. The bow tie model of 21st century palliative care. J Pain Symptom Manage. 2014;47(1):e2-5.
16. Hui D, De La Cruz M, Mori M, et al. Concepts and definitions for "supportive care," "best supportive care," "palliative care," and "hospice care" in the published literature, dictionaries, and textbooks. Supp Care Cancer. 2013;21(3):659-85.
17. Pastrana T, Ostgarthe C, Elsner F, et al. A matter of definition - key elements identified in a discourse analysis of definitions of palliative care. Palliat Med. 2008;22:222-32.
18. Twamley K, Craig F, Kelly P, et al. Underlying barriers to referral to paediatric palliative care services: knowledge and attitudes of health care professionals in a paediatric tertiary care centre in the United Kingdom. J Child Health Care. 2014;18(1):19-30.
19. Together for Short Lives. A Guide to Children's Palliative Care. Bristol: Together for Short Lives; 2018.
20. National Institute for Health and Care Excellence (NICE). End of Life Care for Infants, Children and Young People. NICE Guideline NG61. 2016 https://www.nice.org.uk/guidance/ng61/resources/end-of-life-care-for-infants-children-and-young-people-with-lifelimiting-conditions-planning-and-management-pdf-1837568722885
21. NHS England. E03/S/h - 2013/14 NHS standard contract for paediatric medicine: palliative care particulars, schedule 2 – the services, A - service specifications. NHS England; 2013.

22. Knapp C, Thompson L, Madden V, et al. Paediatricians' perceptions on referrals to paediatric palliative care. Palliat Med. 2009;23(5):418-24.
23. Knapp C, Woodworth L, Wright M, Downing J, Drake R, Fowler-Kerry S, et al. Pediatric Palliative Care Provision Around the World: A Systematic Review. Pediatr Blood Cancer. 2011;57:361-8.
24. Hain R, Devins M, Hastings R, Noyes J. Paediatric palliative care: development and pilot study of a 'Directory' of life-limiting conditions. BMC Palliat Care. 2013;12(1):43.
25. Rempel G. Technological advances in pediatrics: challenges for parents and nurses . J Paed Nurs. 2004;19(1):13-24.
26. Shaw K, Brook L, Mpundu-Kaambwa C, et al. The Spectrum of Children's Palliative Care Needs: a classification framework for children with life-limiting or life-threatening conditions. BMJ Support Palliat Care. 2015;5(3):249-58.
27. Price J, Jordan J, Prior L, et al. Comparing the needs of families of children dying from malignant and non-malignant disease: an in-depth qualitative study. BMJ Support Palliat Care. 2012;2(2):127-32.
28. Collins A, Hennessy-Anderson N, et al. Lived experiences of parents caring for a child with a life-limiting condition in Australia: A qualitative study. Palliat Med. 2016;30(10):950-9.
29. Monterosso L, Kristjanson L, Aoun S, et al. Supportive and palliative care needs of families of children with life-threatening illnesses in Western Australia: evidence to guide the development of a palliative care service. Palliat Med. 2007;21(8):689-96.
30. Baumbusch J, Mayer S, Sloan-Yip I. Alone in a Crowd? Parents of Children with Rare Diseases' Experiences of Navigating the Healthcare System. J Gen Coun 2018 https://doi.org/10.1007/s10897-018-0294-9.
31. Together for Short Lives. Commissioning children's palliative care in England: 2017 edition. Bristol: Together for Short Lives 2017. https://www.togetherforshortlives.org.uk/changing-lives/speaking-up-for-children/policy-advocacy/commissioning-england-2017/
32. Bona K, Wolfe J. Disparities in Pediatric Palliative Care: An Opportunity to Strive for Equity. Pediatrics. 2017;140(4):e20171662. doi: 10.1542/peds.2017-1662.
33. Coad J, Kaur J, Ashley N, et al. Exploring the perceived met and unmet need of life-limited children, young people and families. J Pediatr Nurs. 2015;30(1):45-53.
34. Williams M, Wheeler M. Palliative care: what is it? Home Health Nurs. 2001;19(9):550-6; quiz 7.
35. Mason B, Nanton V, Epiphaniou E, et al. 'My body's falling apart.' Understanding the experiences of patients with advanced multimorbidity to improve care: serial interviews with patients and carers. BMJ Support Palliat Care. 2014.
36. Chen M. Inequity of Palliative Care for Non-Cancer Patients. J Nurs Res 2019;27:1-2.
37. Beringer A, Eaton N, Jones GL. Providing a children's palliative care service in the community through fixed-term grants: the staff perspective. Child Care Health Dev. 2007;33(5):619.
38. Mellor C, Heckford E, Frost J. Developments in paediatric palliative care. Paed Child Health. 2012;22(3):115-20.
39. Friedrichsdorf S, Kang T. The management of pain in children with life-limiting illnesses. Ped Clin N America. 2007;54(5):645-72.
40. MacNamara-Goodger K, Feudtner C. History and epidemiology. In: Goldman A, Hain R, Liben S, editors. Oxford Textbook of Palliative Care for Children 2nd ed. Oxford: Oxford University Press; 2012.
41. Neuberger J. More care, less pathway: a review of the Liverpool Care Pathway. https://www.gov.uk/government/uploads/system/uploads/attachment_data/file/212450/Liverpool_Care_Pathway.pdf; 2013.
42. Parliamentary and Health Service Ombudsman. Dying without Dignity: Investigations by the Parliamentary and Health Service Ombudsman into complaints about end of life care. . London; 2015. https://www.ombudsman.org.uk/publications/dying-without-dignity-0

43.   National Palliative and End of Life Care Partnership. Ambitions for Palliative and End of Life Care: A national framework for local action. http://endoflifecareambitions.org.uk/wp-content/uploads/2015/09/Ambitions-for-Palliative-and-End-of-Life-Care.pdf; 2015.

44.   Office for National Statistics. Deaths registered in England and Wales: 2017. https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarriages/deaths/bulletins/deathsregistrationsummarytables/2017; 2017.

45.   Sidebotham P, Fraser J, Fleming P, et al. Patterns of child death in England and Wales. The Lancet. 2014;384(9946):904-14.

46.   Pearson, G A (Ed) Why Children Die: A Pilot Study 2006; England (South West, North East and West Midlands), Wales and Northern Ireland. London: CEMACH. 2008.

47.   Department of Health. Palliative Care Statistics for Children and Young Adults. London: Department of Health; 2007. https://webarchive.nationalarchives.gov.uk/20130124074133/http://www.dh.gov.uk/prod_consum_dh/groups/dh_digitalassets/@dh/@en/documents/digitalasset/dh_074699.pdf

48.   Goldman A, Beardsmore S, Hunt J. Palliative care for children with cancer - home, hospital or hospice? Arch Dis Child. 1990;65(6):641-3.

49.   Feudtner C, Silvera M, Christakis D. Where do children with complex chronic conditions die? Patterns in Washington State. Pediatrics. 2002;109(4):656-60.

50.   Plunkett A, Parslow RC. Is it taking longer to die in paediatric intensive care in England and Wales? Arch Dis Child. 2016;101:798-802.

51.   Brooten D, Youngblut J, Seagrave L, et al. Parent's perceptions of health care providers actions around child ICU death: What helped, what did not. Am J Hosp Palliat Med. 2013;30(1):40-9.

52.   Prentice T, Janvier A, Gillam L, et al. Moral distress within neonatal and paediatric intensive care units: a systematic review. Arch Dis Child. 2016;101(8):701-8.

53.   Mitchell S, Spry J, Hill E, et al. Parental experiences of end of life care decision-making for children with life-limiting conditions in the paediatric intensive care unit: a qualitative interview study. BMJOpen 2019;9(5):e028548.

54.   BBC News. Charlie Gard: The story of his parents' legal fight https://www.bbc.co.uk/news/health-405544622017

55.   BBC News. Isaiah Haastrup parents announce death of 'brave baby boy' https://www.bbc.co.uk/news/uk-england-london-433281532018

56.   BBC News. Alfie Evans: Legal battle toddler dies https://www.bbc.co.uk/news/uk-439330562018

57.   Staniszewska S, Brett J, Simera I, Seers K, Mockford C, Goodlad S, et al. GRIPP2 reporting checklists: tools to improve reporting of patient and public involvement in research. Br Med J. 2017;358.

58.   INVOLVE. What is public involvement in research? http://www.invo.org.uk/find-out-more/what-is-public-involvement-in-research-2/

59.   Buck D, Gamble C, Dudley L, et al. From plans to actions in patient and public involvement: qualitative study of documented plans and the accounts of researchers and patients sampled from a cohort of clinical trials. BMJ Open. 2014;4(12).

60.   Ennis L, Wykes T. Impact of patient involvement in mental health research: longitudinal study. Br J Psych. 2013;203(5):381-6.

61.   Ocloo J, Matthews R. From tokenism to empowerment: progressing patient and public involvement in healthcare improvement. BMJ Qual Safe. 2016;25(8):626-32.

62.   Coad J, Shaw K. Is children's choice in health care rhetoric or reality? A scoping review. J Adv Nurs. 2008;64(4):318-27.

63.   UNICEF. The United Nations Convention on the Rights of the Child. http://www.unicef.org.uk/Documents/Publication-pdfs/UNCRC_PRESS200910web.pdf; 1990.

64.   Bird D, Culley L, Lakhanpaul M. Why collaborate with children in health research: an analysis of the risks and benefits of collaboration with children. Arch Dis Child Ed & Pract. 2013;98(2):42-8.

65.   RCPCH&Us The voice of children. Children and young people's participation and advocacy resources http://www.rcpch.ac.uk/and-us-resources: Royal College of Paediatrics and Child Health

66.   INVOLVE. Involving Children and Young People http://www.invo.org.uk/find-out-more/how-to-involve-people/involving-children-and-young-people/

67.   Research and Us: Infants', Children's and Young People's Child Health Research Charter. http://www.rcpch.ac.uk/improving-child-health/research-and-surveillance/infants-children-and-young-people%E2%80%99s-research-charte: Royal College of Paediatrics and Child Health 2016

68.   Bailey S, Boddy K, Briscoe S, et al. Involving disabled children and young people as partners in research: a systematic review. Child Care Health Dev. 2015;41(4):505-14.

69.   Royal College of Paediatrics and Child Health. Recipes for Engagement http://www.rcpch.ac.uk/sites/default/files/user31705/%26Us%20Recipes%20for%20Engagement_IF_A5_AW_ONLINE.pdf2016

70.   Barber R, Beresford P, Boote J, et al. Evaluating the impact of service user involvement on research: a prospective case study. Int J Consumer Studies. 2011;35(6):609-15.

71.   Healthtalk.org. Researchers' experiences of patient & public involvement http://www.healthtalk.org/peoples-experiences/medical-research/patient-public-involvement-researchers/overview2016

72.   Bate J, Ranasinghe N, Ling R, et al. Public and patient involvement in paediatric research. Arch Dis Child Ed Pract. 2016. 101(3):158-61. doi: 10.1136/archdischild-2015-309500.

73.   Popay J, Collins M. The Public Involvement Impact Assessment Framework (PiiAF): Executive Summary. University of Lancaster; 2014.

74.   Brett J, Staniszewska S, Mockford C, et al. A Systematic Review of the Impact of Patient and Public Involvement on Service Users, Researchers and Communities. The Patient. 2014;7(4):387-95.

75.   Brett J, Staniszewska S, Mockford C, et al. Mapping the impact of patient and public involvement on health and social care research: a systematic review. Health Expect. 2014;17(5):637-50.

76.   Jamal F, Langford R, Daniels P, et al. Consulting with young people to inform systematic reviews: an example from a review on the effects of schools on health. Health Expect. 2015;18(6):3225-35.

77.   Oliver K, Rees R, Brady L, et al. Broadening public participation in systematic reviews: a case example involving young people in two configurative reviews. Res Synth Methods. 2015;6(2):206-17.

78.   Hunt A, Brown E, Coad J, et al. "Why does it happen like this?' Consulting with users and providers prior to an evaluation of services for children with life limiting conditions and their families. J Child Health Care. 2015;19(3):320-33.

79.   Tume LN, Preston J, Blackwood B. Parents' and young people's involvement in designing a trial of ventilator weaning. Nurs Crit Care. 2016;21(3):e10-e8.

80.   Luchtenberg M, Maeckelberghe E, Locock L, et al. Young People's Experiences of Participation in Clinical Trials: Reasons for Taking Part. Am J Bioethics. 2015;15(11):3-13.

81.   Braun V, Clarke V. Using thematic analysis in psychology. Qual Res Psychology. 2006;3(2):77-101.

82.   Wilson P, Mathie E, Keenan J, et al. Health Services and Delivery Research. ReseArch with Patient and Public invOlvement: a RealisT evaluation - the RAPPORT study. Southampton (UK): NIHR Journals Library

83.   Boote J. Patient and Public Involvement in Health and Social Care Research: A Bibliography. 2011.

84.   Read S, Maslin-Prothero S. The involvement of users and carers in health and social research: the realities of inclusion and engagement. Qual Health Res. 2011;21(5):704-13.

85.   Tullo E, Robinson L, Newton J. Comparing the perceptions of academics and members of the public about patient and public involvement in ageing research. Age Ageing. 2015;44(3):533-6.

86. Weil L, Lemer C, Webb E, et al. The voices of children and young people in health: where are we now? Arch Dis Child. 2015;100(10):915-7. doi: 10.1136/archdischild-2014-307492.

87. Rahimzadeh V, Bartlett G, Longo C, et al. Promoting an ethic of engagement in pediatric palliative care research. BMC Palliat Care. 2015;14(1):50.

88. Mahmood-Yousuf K, Munday D, King N, et al. Interprofessional relationships and communication in primary palliative care: impact of the Gold Standards Framework. Br J Gen Pract. 2008;58(549):256-63.

89. Walshe C, Luker KA. District nurses' role in palliative care provision: A realist review. Int J Nurs Stud. 2010;47(9):1167-83.

90. Bliss J, While AE. Meeting the needs of vulnerable patients: The need for team working across general practice and community nursing services. London J Prim Care (Abingdon). 2014;6(6):149-53.

91. Centre for Reviews and Dissemination. Systematic Reviews: CRD's guidance for undertaking reviews in health care. University of York, 2008.

92. Higgins P, Green S, Ed. The Cochrane Collaboration. Cochrane Handbook for Systematic Reviews of Interventions. England Wiley-Blackwell; 2011.

93. Shamseer L, Moher D, Clarke M, et al. Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P) 2015: elaboration and explanation. Br Med J. 2015;349:g7647.

94. Methley AM, Campbell S, Chew-Graham C, McNally R, Cheraghi-Sohi S. PICO, PICOS and SPIDER: a comparison study of specificity and sensitivity in three search tools for qualitative systematic reviews. BMC Health Serv Res. 2014;14:579-

95. Popay J, Roberts H, Sowden A, et al. Guidance on the conduct of narrative synthesis in systematic reviews. Lancaster: ESRC Research Methods Programme; 2006.

96. Friedrichsdorf S, Postier A, Dreyfus J, et al. Improved Quality of Life at End of Life Related to Home-Based Palliative Care in Children with Cancer. J Palliat Med. 2015;18(2):143-50.

97. Niswander LM, Cromwell P, Chirico J, Gupton A, Korones DN. End-of-life care for children enrolled in a community-based pediatric palliative care program. J Palliat Med. 2014;17(5):589-91.

98. Sheetz MJ, Bowman MAS. Parents' Perceptions of a Pediatric Palliative Program. Am J Hosp Palliat Med. 2013;30(3):291-6.

99. Postier A, Chrastek J, Nugent S, et al. Exposure to Home-Based Pediatric Palliative and Hospice Care and Its Impact on Hospital and Emergency Care Charges at a Single Institution. J Palliat Med. 2014;17(2):183-8.

100. Fraser L, van Laar M, Miller M, et al. Does referral to specialist paediatric palliative care services reduce hospital admissions in oncology patients at the end of life? Br J Cancer. 2013;108(6):1273-9.

101. Groh G, Borasio GD, Nickolay C, et al. Specialized Pediatric Palliative Home Care: A Prospective Evaluation. J Palliat Med. 2013;16(12):1588-94.

102. Nolte-Buchholz S, Zernikow B, Wager J. Pediatric Patients Receiving Specialized Palliative Home Care According to German Law: A Prospective Multicenter Cohort Study. Children (Basel). 2018;5(6):66. Published 2018 May 31. doi:10.3390/children5060066

103. Zernikow B, Szybalski K, Hubner-Mohler B, et al. Specialized pediatric palliative care services for children dying from cancer: A repeated cohort study on the developments of symptom management and quality of care over a 10-year period. Palliat Med. 2019;33(3):381-91.

104. Herbert A, Bradford N, Donovan L, et al. Development of a state-wide pediatric palliative care service in Australia: Referral and outcomes over two years. J Palliat Med. 2014;17(3):288-95.

105. Kassam A, Skiadaresis J, Alexander S, et al. Differences in end-of-life communication for children with advanced cancer who were referred to a palliative care team. Pediatric Blood & Cancer. 2015;62(8):1409-13.

106. Widger K, Sutradhar R, Rapoport A, Vadeboncoeur C, Zelcer S, Kassam A, et al. Predictors of Specialized Pediatric Palliative Care Involvement and Impact on Patterns of End-of-Life Care in Children With Cancer. J Clin Oncol. 2018:JCO2017756312.

107. Chong PH, De Castro Molina JA, Teo K, Tan WS. Paediatric palliative care improves patient outcomes and reduces healthcare costs: evaluation of a home-based program. BMC Palliat Care. 2018;17(1):11.

108. Critical Appraisal Skills Programme: CASP checklists. http://www.casp-uk.net/casp-tools-checklists

109. Junger S, Hasan C, Burger A, et al. Coordination of paediatric palliative home care services. Palliat Med. 2010;1):S22.

110. Wee B, Hadley G, Derry S. How useful are systematic reviews for informing care practice? Survey of 25 Cochrane systematic reviews. BMC Palliat Care. 2008;7(13).

111. Crocker JC, Beecham E, Kelly P, et al. Inviting parents to take part in paediatric palliative care research: A mixed-methods examination of selection bias. Palliat Med. 2015;29(3):231-40.

112. National Hospice and Palliative Care Organisation (NHPCO')s Standared for Pediatric Care. https://www.nhpco.org/quality/nhpco%E2%80%99s-standards-pediatric-care

113. European Association for Palliative Care. IMPaCCT: standards for paediatric palliative care in Europe. Eur J Palliat Care. 2007;14(3):109-14.

114. Bluebond-Langner M, Beecham E, Candy B, et al. Preferred place of death for children and young people with life-limiting and life-threatening conditions: a systematic review of the literature and recommendations for future inquiry and policy. Palliat Med. 2013;27(8):705-13.

115. Wolfe J, Grier HE, Klar N, et al. Symptoms and Suffering at the End of Life in Children with Cancer. New Eng J Med. 2000;342(5):326-33.

116. Ullrich C, Morrison RS. Pediatric palliative care research comes of age: what we stand to learn from children with life-threatening illness. J Palliat Med. 2013;16(4):334–336. doi:10.1089/jpm.2013.9518

117. Breen M, Anderson A-K, Jones KP. Evaluation of Place of Care at Time of Death at One United Kingdom Principal Treatment Centre for Children with Cancer. Europ J Onc Nurs. 2011;15(3):277.

118. Sastry J, Ronghe M, Murphy D, Hall A, Belmore J, Marshall K, et al. Nurse led palliative care at home for children with brain tumours-west of Scotland experience. Neuro-Oncology. 2014;16:i109.

119. Mastro K, Johnson J, McElvery N, et al. The benefits of a nurse-driven patient- and family-centred pediatric palliative care program. J Nurs Admin. 2015;45(9):423-8.

120. Martinson I, Armstrong G, Geis DP, et al. Home Care for Children Dying of Cancer. Pediatr. 1978;62(1):106-13.

121. Van Der Tempel A, Groenenberg AP, Vallianatos S. Case management in paediatric palliative care. Arch Dis Child. 2014;99:A536-A7.

122. Academy of Medical Royal Colleges. The benefits of consultant-delivered care. London: Academy of Medical Royal Colleges. 2012.

123. NHS England. Delivering the Forward View: NHS planning guidance 2016/17 - 2020/21. https://www.england.nhs.uk/wp-content/uploads/2015/12/planning-guid-16-17-20-21.pdf

124. Morstad-Boldt A, Yusuf F, Himelstein B. Perceptions of the term palliative care. J Palliat Med. 2006;9(5):1128-36.

125. Fraser J, Harris N, Berringer AJ, et al. Advanced care planning in children with life-limiting conditions - The Wishes Document. Arch Dis Child. 2010;95(2):79-82.

126. Pawson R, Tilley N. Realistic evaluation. London: Sage Publications; 1997.

127. Pawson R, Greenhalgh T, Harvey G, et al. Realist review--a new method of systematic review designed for complex policy interventions. J Health Serv Res Policy. 2005;10 Suppl 1:21-34.

128. Papoutsi C, Mattick K, Pearson M, et al. Health Services and Delivery Research. Interventions to improve antimicrobial prescribing of doctors in training (IMPACT): a realist review. Southampton (UK): NIHR Journals Library. 2018.

129. Wong G, Greenhalgh T, Westhorp G, et al. RAMESES publication standards: realist syntheses. BMC Med. 2013;11(1):21.
130. Ford J, Wong G, Jones A, et al. Access to primary care for socioeconomically disadvantaged older people in rural areas: a realist review. BMJ Open. 2016;6(5).
131. Weetman K, Wong G, Scott E, et al. Improving best practice for patients receiving hospital discharge letters: a realist review protocol. BMJ Open. 2017;7(11).
132. Neil L, Clarke S. Learning to live with childhood cancer: a literature review of the parental perspective. Int J Palliat Nurs. 2010;16(3):110-9.
133. Heinze K, Nolan M. Parental decision making for children with cancer at the end of life: a meta-ethnography. J Pediatr Oncol Nurs. 2012;29(6):337-45.
134. Wang J, Kearney J. The experience of Chinese American parents of children with life-limiting illness: A comprehensive review. Int J Palliat Nurs. 2013;19(7):347-54.
135. Melin-Johansson C, Axelsson I, Jonsson Grundberg M, When a Child Dies: Parents' Experiences of Palliative Care - An Integrative Literature Review. J Pediatr Nurs. 2014;29(6):660.
136. Montgomery K, Sawin K, Hendricks-Ferguson V. Experiences of pediatric oncology patients and their parents at end of life: A systematic review. J Pediatr Onc Nurs. 2016;33(2):85-104.
137. Darnill S, Gamage B. The patient's journey: palliative care--a parent's view. Br Med J. 2006;332(7556):1494-5.
138. Nimmo S. Letting my daughter go. Br Med J. 2018;360.
139. O'Shea E, Kanarek R. Understanding pediatric palliative care: What it is and what it should be. J Pediatr Onc Nurs. 2013;30(1):34-44.
140. Kavas MV. How to Increase the Quality of a Suffering Experience: Lessons Derived From the Diary Narratives of a Dying Adolescent Girl. Omega. 2018;76(3):256-95.
141. Wolfe J, Klar N, Grier HE, et al. Understanding of prognosis among parents of children who died of cancer: impact on treatment goals and integration of palliative care. J Am Med Assoc. 2000;284(19):2469-75.
142. Surkan P, Kreicbergs U, Valdimarsdottir U, et al. Perceptions of inadequate health care and feelings of guilt in parents after the death of a child to a malignancy: a population-based long-term follow-up. J Palliat Med. 2006;9(2):317-31.
143. Hechler T, Blankenburg M, Friedrichsdorf SJ, et al. Parents' perspective on symptoms, quality of life, characteristics of death and end-of-life decisions for children dying from cancer. Klin Padiatr. 2008;220(3):166-74.
144. Dighe M, Jadhav S, Muckaden M, et al. Parental concerns in children requiring palliative care. Indian J Palliat Care. 2008;14(1):16-22.
145. Monterosso L, Kristjanson L. Supportive and palliative care needs of families of children who die from cancer: an Australian study. Palliat Med. 2008;22(1):59-69.
146. Monterosso L, Kristjanson L, Phillips M. The supportive and palliative care needs of Australian families of children who die from cancer. Palliat Med. 2009;23(6):526-36.
147. Zelcer S, Cataudella D, Cairney A et al. Palliative care of children with brain tumors: a parental perspective. Arch Pediatr Adolesc Med. 2010;164(3):225-30.
148. Miedema B, Hamilton R, Fortin P,et al. 'You can only take so much, and it took everything out of me': Coping strategies used by parents of children with cancer. Palliat Support Care. 2010;8(2):197-206.
149. Tomlinson D, Hendershot E, Bartels U, et al. Concordance between couples reporting their child's quality of life and their decision making in pediatric oncology palliative care. J Pediatr Onc Nurs. 2011;28(6):319-25.
150. von Lutzau P, Otto M, Hechler T, et al. Children dying from cancer: parents' perspectives on symptoms, quality of life, characteristics of death, and end-of-life decisions. J Palliat Care. 2012;28(4):274-81.
151. Steele A, Kaal J, Thompson A, et al. Bereaved Parents and Siblings Offer Advice to Healthcare Providers and Researchers. J Pediatr Hem Onc. 2013;35(4):253-9.

152. Kars M, Grypdonck M, de Bock L, et al. The parents' ability to attend to the "voice of their child" with incurable cancer during the palliative phase. Health Psychol. 2015;34(4):446-52.
153. Bjork M, Sundler AJ, Hallstrom I, Hammarlund K. Like being covered in a wet and dark blanket - Parents' lived experiences of losing a child to cancer. Europ J Onc Nurs. 2016;25:40-5.
154. Jalmsell L, Lovgren M, Kreicbergs U, et al. Children with cancer share their views: Tell the truth but leave room for hope. Acta Paediatrica. 2016;105(9):1094-9.
155. Nelson M, Kelly D, McAndrew R, Smith P. 'Just gripping my heart and squeezing': Naming and explaining the emotional experience of receiving bad news in the paediatric oncology setting. Patient Educ Couns. 2017;100(9):1751-7.
156. Steele R, Davies B. Impact on parents when a child has a progressive, life-threatening illness. Int J Palliat Nurs. 2006;12(12):576-85.
157. Rempel G, Ravindran V, Rogers L et al. Parenting under pressure: a grounded theory of parenting young children with life-threatening congenital heart disease. J Adv Nurs. 2013;69(3):619-30.
158. Steele R, Siden H, Cadell S, et al. Charting the territory: symptoms and functional assessment in children with progressive, non-curable conditions. Arch Dis Child. 2014;99(8):754-62.
159. Blume E, Balkin E, Aiyagari R, et al. Parental perspectives on suffering and quality of life at end-of-life in children with advanced heart disease: an exploratory study. Pediatr Crit Care Med. 2014;15(4):336-42.
160. Contro N, Larson J, Scofield S, et al. Family perspectives on the quality of pediatric palliative care. Arch Pediatr Adolesc Med. 2002;156(1):14-9.
161. Macdonald M, Liben S, Carnevale F, et al. Parental perspectives on hospital staff members' acts of kindness and commemoration after a child's death. Pediatrics. 2005;116(4):884-90.
162. Hsiao J, Evan E, Zeltzer L. Parent and child perspectives on physician communication in pediatric palliative care. Palliat Support Care. 2007;5(4):355-.
163. Hendricks-Ferguson V. Parental perspectives of initial end-of-life care communication. Int J Palliat Nurs. 2007;13(11):522-31.
164. Konrad Cohen K. Mothers' perspectives on qualities of care in their relationships with health care professionals: The influence of relational and communicative competencies. J Soc Work End Life Palliat Care. 2008;4(1):38-56.
165. Knapp C, Madden V, Wang H, et al. Factors affecting decisional conflict for parents with children enrolled in a paediatric palliative care programme. Int J Palliat Nurs. 2010;16(11):542-7.
166. Contro N, Davies B, Larson J, et al. Away from home: Experiences of Mexican American families in pediatric palliative care. J Soc Work End Life Palliat Care. 2010;6(3-4):185-204.
167. Menezes A. Moments of realization: life-limiting illness in childhood-perspectives of children, young people and families. Int J Palliat Nurs. 2010;16(1):41-7.
168. Knapp C, Madden V, Curtis C, et al. Family Support in Pediatric Palliative Care: How Are Families Impacted by their Children's Illnesses? J Palliat Med. 2010;13(4):421-6.
169. Fauman K, Pituch K, Han Y, et al. Predictors of depressive symptoms in parents of chronically ill children admitted to the pediatric intensive care unit. Am J Hosp Palliat Med. 2011;28(8):556-63.
170. Kirk S, Pritchard E. An exploration of parents' and young people's perspectives of hospice support. Child Care Health Dev. 2012;38(1):32-40.
171. Robert R, Zhukovsky D, Mauricio R, et al. Bereaved parents' perspectives on pediatric palliative care. J Soc Work End Life Palliat Care. 2012;8(4):316-38.
172. Gupta V, Prescott H. "That must be so hard"- Examining the impact of children's palliative care services on the psychological well-being of parents. Clinical Child Psychology Psychiatry. 2013;18(1):91-9.
173. Gilmer M, Foster T, Bell C, et al. Parental perceptions of care of children at end of life. Am J Hosp Palliat Med. 2013;30(1):53-8.
174. Gaab E, Glynn Owens R, Macleod R. Primary caregivers decisions around communicating about death with children involved in pediatric palliative care. J Hosp Palliat Nurs. 2013;15(6):322-9.

175. Hill DL, Miller VA, Hexem KR, et al. Problems and hopes perceived by mothers, fathers and physicians of children receiving palliative care. Health Expect. 2015;18(5):1052-65.

176. Sullivan J, Monagle P, Gillam L. What parents want from doctors in end-of-life decision-making for children. Arch Dis Child. 2014;99(3):216-20.

177. Gaab E, Owens G, MacLeod R. Siblings caring for and about pediatric palliative care patients. J Palliat Med. 2014;17(1):62-7.

178. van der Geest I, van den Heuvel-Eibrink M, Falkenburg N, et al. Parents' Faith and Hope during the Pediatric Palliative Phase and the Association with Long-Term Parental Adjustment. J Palliat Med. 2015;18(5):402-7.

179. van der Geest I, Darlington A, Streng I, et al. Parents' experiences of pediatric palliative care and the impact on long-term parental grief. J Pain Symptom Manage. 2014;47(6):1043-53.

180. Rodriguez A, King N. Sharing the care: the key-working experiences of professionals and the parents of life-limited children. Int J Palliat Nurs. 2014;20(4):165-71.

181. Gans D, Hadler MW, Chen X, Wu S-H, Dimand R, Abramson JM, et al. Impact of a Pediatric Palliative Care Program on the Caregiver Experience. J Hosp Palliat Nurs. 2015;17(6):559-65.

182. Kuan G, Low W. Parents' perspectives on the important aspects of care in children dying from life limiting conditions: A qualitative study. Med J Malaysia. 2015;70(5):295-9.

183. Zimmermann K, Bergstraesser E, Engberg S, et al. When parents face the death of their child: a nationwide cross-sectional survey of parental perspectives on their child's end-of life care. BMC Palliat Care. 2016;15:30.

184. Kirk S, Glendinning C. Supporting 'expert' parents—professional support and families caring for a child with complex health care needs in the community. Int J Nurs Studies. 2002;39(6):625-35.

185. Konrad Cohen S. What Parents of Seriously Ill Children Value: Parent-to-Parent Connection, and Mentorship. Omega. 2007;55(2):117-30.

186. Dussel V, Kreicbergs U, Hilden JM, et al. Looking Beyond Where Children Die: Determinants and Effects of Planning a Child's Location of Death. J Pain Sympt Manage. 2009;37(1):33-43.

187. Surkan P, Dickman PW, Steineck G, et al. Home care of a child dying of a malignancy and parental awareness of a child's impending death. Palliat Med. 2006;20(3):161-9.

188. Rycroft-Malone J, McCormack B, Hutchinson AM, et al. Realist synthesis: illustrating the method for implementation research. Implementation Science. 2012;7(1):33.

189. Hynson J, Aroni R, Bauld C,et al. Research with bereaved parents: a question of how not why. Palliat Med. 2006;20(8):805-11.

190. Lefeubvre C, Viallard ML, Schell M. Pediatric palliative care: A national survey of French pediatric residents' knowledge, education, and clinical experience. Arch De Pediatr. 2014;21(8):834-44.

191. Rusalen F, Ferrante A, Po C, et al. Pain therapy, pediatric palliative care and end-of-life care: training, experience, and reactions of pediatric residents in Italy. Europ J Pediatr. 2014;173(9):1201-7.

192. Sheetz M, Bowman M. Pediatric palliative care: an assessment of physicians' confidence in skills, desire for training, and willingness to refer for end-of-life care. Am J Hosp Palliat Care. 2008;25(2):100-5.

193. Straatman L, Miller T. Paediatric palliative care: a survey of paediatricians and family practitioners. BMJ Support Palliat Care. 2013;3(3):366-71.

194. Boyd K, Moine S, Murray SA, et al. Should palliative care be rebranded? 2019;364:l881.

195. Wu KL, Friderici J, Goff SL. The Impact of a Palliative Care Team on Residents' Experiences and Comfort Levels with Pediatric Palliative Care. J Palliat Med. 2014;17(1):80-4.

196. National Institute for Health and Care Excellence (NICE). End of Life care for infants, children and young people. Quality Standard [QS160] https://www.nice.org.uk/guidance/qs1602017

197. Sackett DL. Evidence-based medicine. Semin Perinat. 1997; 21(1):3-5.

198. Sackett DL, Rosenberg W, Gray J,et al. Evidence based medicine: what it is and what it isn't. Br Med J. 1996;312(7023):71-2.

199. Evidence-based medicine working group. Evidence-based medicine. A new approach to teaching the practice of medicine. JAMA. 1992;268(17):2420-5.
200. Evans D. Hierarchy of evidence: a framework for ranking evidence evaluating healthcare interventions. J Clin Nurs. 2003;12(1):77-84.
201. Murad MH, Asi N, Alsawas M, et al. New evidence pyramid. 2016;21(4):125-7.
202. Greenhalgh T, Howick J, Maskrey N. Evidence based medicine: a movement in crisis? 2014;348.
203. World Palliative Care Alliance, World Health Organisation (2014) Global Atlas of Palliative Care.
204. Addington-Hall J. Research sensitivities to palliative care patients. Eur J Cancer Care (Engl). 2002;11(3):220-4.
205. de Raeve L. Ethical issues in palliative care research. Palliat Med. 1994;8(4):298-305.
206. Sivell S, Prout H, Hopewell-Kelly N, Baillie J, Byrne A, Edwards M, et al. Considerations and recommendations for conducting qualitative research interviews with palliative and end-of-life care patients in the home setting: a consensus paper. BMJ Support Palliat Care. 2019; 9(1):e14-e.
207. Hoare S, Morris ZS, Kelly MP, et al. Do Patients Want to Die at Home? A Systematic Review of the UK Literature, Focused on Missing Preferences for Place of Death. PloSOne. 2015;10(11):e0142723-e.
208. Dzingina M, Higginson I, McCrone P, et al. Development of a Patient-Reported Palliative Care-Specific Health Classification System: The POS-E. Patient. 2017;10(3):353-65.
209. Coombes L, Wiseman T, Lucas G, et al. Health-related quality-of-life outcome measures in paediatric palliative care: A systematic review of psychometric properties and feasibility of use. Palliat Med. 2016;30(10):935-49.
210. The RAMESES Projects Resources and training materials for realist evaluation. http://www.ramesesproject.org/Standards_and_Training_materials.php
211. Emmel N, Greenhalgh J, Manzano A, et al Doing Realist Research. London: SAGE; 2018.
212. Higginson I, Bausewein C, Reilly C, et al. An integrated palliative and respiratory care service for patients with advanced disease and refractory breathlessness: a randomised controlled trial. Lancet Respir Med. 2014;2(12):979-87.
213. Temel J, Greer JA El-Jawahri A, et al. Effects of Early Integrated Palliative Care in Patients With Lung and GI Cancer: A Randomized Clinical Trial. J Clin Oncol. 2017;35(8):834-41.
214. Gomes B, Calanzani N, Curiale V, et al. Effectiveness and cost-effectiveness of home palliative care services for adults with advanced illness and their caregivers. The Cochrane Database of Systematic Reviews. 2013(6):1-279.
215. Higginson I, McCrone P, Hart SR, et al. Is short-term palliative care cost-effective in multiple sclerosis? A randomized phase II trial. J Pain Symptom Manage. 2009;38(6):816-26.
216. Detering K, Hancock A, Reade M, et al. The impact of advance care planning on end of life care in elderly patients: randomised controlled trial. Br Med J. 2010;340.
217. Rosenberg J, Albrecht J, Fromme E, et al. Antimicrobial use for symptom management in patients receiving hospice and palliative care: a systematic review. J Palliat Med. 2013;16(12):1568-74.
218. Jennings A-L, Davies A, Higgins J, et al. A systematic review of the use of opioids in the management of dyspnoea. Thorax. 2002;57(11):939-44.
219. Braun V, Clarke V. Successful Qualitative Research. London: SAGE; 2013.
220. Glaser BG, Strauss AL. The Discovery of Grounded Theory: Strategies for qualitative research. Mill Valley: CA: Sociology Press; 1967.
221. Pawson R. Evidence-Based Policy: A Realist Perspective. London: SAGE; 2006.
222. Popper K. Conjectures and Refutations: the Growth of Scientific Knowledge. London: Routledge; 1963.
223. Popper K. The Logic of Scientific Discovery. 5th ed. London: Routledge Classics; 1992.
224. Bhaskar R. A realist theory of science. Brighton: Harvester; 1975.
225. Merton R. Social Theory and Social Structure. New York: Free Press; 1968.

226. Campbell D. Methodolgy and Epistemology for Social Science: Collected Papers Overman S, editor. Chicago: University of Chicago Press; 1988.
227. Pawson R. The Science of Evaluation: A Realist Manifesto. Los Angeles, CA and London.: SAGE; 2013.
228. Plsek PE, Greenhalgh T. The challenge of complexity in health care. 2001;323(7313):625-8.
229. Greenhalgh T, Papoutsi C. Studying complexity in health services research: desperately seeking an overdue paradigm shift. BMC Med. 2018;16(1):95.
230. Bronfenbrenner U. The bioecological theory of human development. In: Bronfenbrenner U., editor. Making human beings human:Bioecological perspectives on human development. Thousand Oaks, CA: SAGE; 2005. p. 3-15.
231. Trudge J., Mokrova I., Hatfield B., Karnik R. Uses and Misuses of Bronfenbrenner's Bioecological Theory of Human Development. J Fam Theory and Review. 2009;1:198-210.
232. Balint M. The Doctor, his patient and the illness. The Lancet. 1955;265(6866):683-8.
233. Curran J. The Doctor, his Patient and the Illness. Br Med J. 2007;335(7626):941.
234. Petrova M, Dale J, Fulford B. Values-based practice in primary care: easing the tensions between individual values, ethical principles and best evidence. Br J Gen Pract: 2006;56(530):703-9.
235. Office for National Statistics. 2011 Census: Population Estimates for the United Kingdom, March 2011. https://www.ons.gov.uk/census/2011census
236. Jackson S, Jones A, Wiltshire J. Palliative and End of Life Care Report for Children and Young People in the West Midlands. Comissioned by NHS England. https://www.strategyunitwm.nhs.uk/sites/default/files/2018-04/Final_PPC_Report%20V3_2.pdf: The Strategy Unit; 2017.
237. Wyatt S, Bennett A. Palliative and End of Life Care in the West Midlands. A Report for Sustainability and Transformation Partnerships commissioned by NHS England. https://www.strategyunitwm.nhs.uk/sites/default/files/2017-11/Palliative%20and%20End%20of%20Life%20Care%20in%20the%20West%20Midlands%20-%20final%20-%20171005_3.pdf: The Strategy Unit; 2017.
238. Hunt A, Coad, J, West, E, et al. The Big Study for Life-limited Children and their Families: Final research report. Bristol: Together for Short Lives; 2013.
239. Flanagan SM, Greenfield S, Coad J, et al. An exploration of the data collection methods utilised with children, teenagers and young people (CTYPs). BMC Res Notes. 2015;8:61.
240. Charmaz K. Constructing Grounded Theory: A practical guide through qualitative analysis. Nurs Researcher. 2006;13(4):84.
241. Chun Tie Y, Birks M, Francis K. Grounded theory research: A design framework for novice researchers. SAGE Open Med. 2019;7:2050312118822927-.
242. Akard T, Gilmer M, Friedman D, et al. From Qualitative Work to Intervention Development in Pediatric Oncology Palliative Care Research. J Ped Onc Nurs. 2013;30(3):153-60.
243. Kendall M, Harris F, Boyd K, et al. Key challenges and ways forward in researching the "good death": qualitative in-depth interview and focus group study. Br Med J. 2007;334(7592):521.
244. Murray SA, Kendall M, Carduff E, et al. Use of serial qualitative interviews to understand patients' evolving experiences and needs. Br Med J. 2009;339.
245. Pessin H, Galietta M, Nelson C, et al. Burden and benefit of psychosocial research at the end of life. J Palliat Med. 2008;11(4):627-32.
246. Tomlinson D, Bartels U, Hendershot E et al. Challenges to participation in paediatric palliative care research: a review of the literature. Palliat Med. 2007;21(5):435-40.
247. Kars MC, van Thiel GJ, van der Graaf R, et al. A systematic review of reasons for gatekeeping in palliative care research. Palliat Med. 2016;30(6):533-48.
248. Neighbour R. The Inner Consultation. Berkshire, UK: LibraPharm; 1987.
249. Carter B, Coad J, Goodenough T. Community Children's Nursing in England: An appreciative review of CCNs. Department of Health Report. NHS at Home: Community Children's Nursing Services.

http://www.dh.gov.uk/en/Publicationsandstatistics/Publications/PublicationsPolicyAndGuidance/DH_1248-98. 2009.

250. Bellinger A, Elliott T. What Are You Looking At? The Potential of Appreciative Inquiry as a Research Approach for Social Work. Br J Soc Work. 2011;41(4):708-25.

251. Angell C, Alexander J, Hunt J. 'Draw, write and tell': A literature review and methodological development on the 'draw and write' research method. J Early Child Res. 2015;13(1):17-28. https://doi.org/10.1177/1476718X14538592

252. Greene S., Hill M. Researching Children's Experience: Methods and Methodological Issues. In: Greene S., Hogan D., editors. Researching Children's Experience. London: SAGE; 2005. p. 8-9.

253. Greig A, Taylor J, MacKay T. Doing Research with Children. A practical guide. London: SAGE; 2008. p. 113-20.

254. University of Oxdord Nuffield Department of Primary Care Health Sciences. Introduction to Analyising Qualitative Interviews https://www.phc.ox.ac.uk/study/short-courses-in-qualitative-research-methods/analysing-qualitative-interviews

255. Burnard P, Gill P, Stewart K, et al. Analysing and presenting qualitative data. Br Dent J. 2008;204(8):429-32.

256. Bingley A, Thomas C, Brown J, et al. Developing narrative research in supportive and palliative care: the focus on illness narratives. Palliat Med. 2008;22(5):653-8.

257. Lieblich A, Tuval-Mashiach R, Zilber T. Narrative research: reading, analysis and interpretation. London: Sage; 1998.

258. Wong G., Westhorp G, Manzano A. et al. RAMESES II reporting standards for realist evaluations. BMC Med. 2016;14(96).

259. Medical Research Council. MRC Ethics Guide: Medical research involving children. London. www.mrc.ac.uk: Medical Research Council. 2004.

260. Shaw C, Brady L-M, Davey C. Guidelines for Research with Children and Young People. London: National Children's Bureau; 2011.

261. Calman L, Brunton L, Molassiotis A. Developing longitudinal qualitative designs: lessons learned and recommendations for health services research. BMC Med Res Method. 2013;13(1):1-10.

262. Bluebond-Langner M, Belasco J, Wander M. "I want to live until I don't want to live anymore". Involving children with life-threatening and life-shortening illnesses in decision making about care and treatment. Nurs Clin N Am. 2010;45:329-43.

263. Greenhalgh T, Papoutsi C. Spreading and scaling up innovation and improvement. Br Med J. 2019;365:l2068.

264. Nadeem S. Personal communication. 2016.

265. Namisango E, Bristowe K, Allsop M, et al. Symptoms and Concerns Among Children and Young People with Life-Limiting and Life-Threatening Conditions: A Systematic Review Highlighting Meaningful Health Outcomes. Patient. 2019;12(1):15-55.

266. Smith J, Cheater F, Bekker H. Parents' experiences of living with a child with a long-term condition: a rapid structured review of the literature. Health Expect. 2015;18(4):452-74.

267. Borgstrom E, Walter T. Choice and compassion at the end of life: A critical analysis of recent English policy discourse. Soc Sci Med. 2015;136-137:99-105.

268. Gilljam B-M, Arvidsson S, Nygren JM, et al. Promoting participation in healthcare situations for children with JIA: a grounded theory study. Int J Qual Studies on Health and Well-being. 2016;11(1):30518.

269. Coyne I. Giving children a voice: Investigation of children's experiences of participation in consultation and decision-making in Irish hospitals. 2006. Giving Children a Voice . Government Publications Stationary Office Dublin, Dublin. ISBN 978-0755716623

270. Coyne I, Kirwan L. Ascertaining children's wishes and feelings about hospital life. J Child Health Care. 2012;16(3):293-304.

271. Mårtenson E, Fägerskiöld A. A review of children's decision-making competence in health care. J Clin Nurs 2008;17(23):3131-41

272. Picker Institute. Principles of patient centred care https://nexusipe.org/informing/resource-center/picker-institute%E2%80%99s-eight-principles-patient-centered-care.2015

273. Ahmad N. Ellins J, Krelle H, et al. Person-centred care: from ideas to action. Bringing together the evidence on shared decision making and self-management support. London: The Health Foundation; 2014.

274. Marshall M, Bibby J. Supporting patients to make the best decisions. Br Med J 2011;342:d2117. doi: 10.1136/bmj.d2117

275. Royal College of General Practitioners (RCGP). An inquiry into Patient Centred Care in the 21st Century. Implications for general practice and primary care. London: RCGP; 2014.

276. Royal College of Paediatrics and Child Health (RCPCH). Inquiry into Patient Centred Care in the 21st Century. Response from the Royal College of Paediatrics and Child Health. London: RCPCH; 2014.

277. Dewan T, Cohen E. Children with medical complexity in Canada. Paediatr Child Health. 2013;18(10):518-22.

278. Uygur J, Brown JB, Herbert C. Understanding compassion in family medicine: a qualitative study. 2019;69(680):e208-e16.

279. Gulliford M, Naithani S, Morgan M. What is 'continuity of care'? J Health Serv Res Policy. 2006;11(4):248-50.

280. Pereira Gray D, Sidaway-Lee K, White E, et al. Continuity of care with doctors—a matter of life and death? A systematic review of continuity of care and mortality. BMJOpen 2018;8(6):e021161.

281. Lotz JD, Daxer M, Jox RJ, Borasio GD, Fuhrer M. "Hope for the best, prepare for the worst": A qualitative interview study on parents' needs and fears in pediatric advance care planning. Palliat Med. 2017;31(8):764-71.

282. Lotz J, Jox R, Borasio G, et al. Pediatric advance care planning: a systematic review. Pediatri. 2013;131(3):e873-80.

283. Petrova M, Riley J, Abel J, et al. Crash course in EPaCCS (Electronic Palliative Care Coordination Systems): 8 years of successes and failures in patient data sharing to learn from. BMJ Support Palliat Care. 2018;8(4):447-55.

284. Rimmer A. Increase paediatric trainee numbers to meet workforce shortfall, says Royal College. Br Med J. 2017;357:j2075.

285. Connor S, Downing J, Marston J et al. Estimating the global need for palliative care for children: a cross-sectional analysis. J Pain Sympt Manage. 2017;53(2):171-7.

286. Ekberg S, Danby S, Rendle-Short J, et al. Discussing death: Making end of life implicit or explicit in paediatric palliative care consultations. Patient Ed Couns. 2019;102(2):198-206.

287. Levine D, Mandrell B, Sykes A, et al. Patients' and Parents' Needs, Attitudes, and Perceptions About Early Palliative Care Integration in Pediatric Oncology. JAMA oncology. 2017;3(9):1214-20.

288. Doorenbos A, Starks H, Bourget E, et al. Examining Palliative Care Team Involvement in Automatic Consultations for Children on Extracorporeal Life Support in the Pediatric Intensive Care Unit. J Palliat Med. 2013;16(5):492-5.

289. Bergstraesser E. Pediatric palliative care: a reflection on terminology. Palliat Care. 2013;7:31-6.

290. Fowler K, Poehling K, Billheimer D, Hamilton R, Wu H, Mulder J, et al. Hospice referral practices for children with cancer: a survey of pediatric oncologists. J Clin Oncol. 2006;24(7):1099-104.

291. Fulford KWM. Values-based practice: a new partner to evidence-based practice and a first for psychiatry? Mens Sana Monogr. 2008;6(1):10-21.

292. Brown M, Brown G. Evidence-based to value-based medicine: Amer Medical Assn; 2005.

293. May C, Finch T. Implementing, Embedding, and Integrating Practices: An Outline of Normalization Process Theory. Sociology. 2009;43(3):535-54.

294. McEvoy R, Ballini L, Maltoni S, et al. A qualitative systematic review of studies using the normalization process theory to research implementation processes. Implement Sci. 2014;9:2-.

295. Murray E, Treweek S, Pope C, MacFarlane A, Ballini L, Dowrick C, et al. Normalisation process theory: a framework for developing, evaluating and implementing complex interventions. BMC Med. 2010;8(1):63.

296. Villanueva G, Murphy MS, Vickers D, et al. End of life care for infants, children and young people with life limiting conditions: summary of NICE guidance. Br Med J. 2016;355.

297. Sleeman KE, de Brito M, Etkind S, et al. The escalating global burden of serious health-related suffering: projections to 2060 by world regions, age groups, and health conditions. The Lancet Global Health. 2019: 7(7):e883-e892. doi: 10.1016/S2214-109X(19)30172-X.

298. Laddie J, Craig F, Brierley J, et al. Withdrawal of ventilatory support outside the intensive care unit: guidance for practice. Arch Dis Child. 2014;99(9):812-6.

299. Larcher V, Craig F, Bhogal K, Wilkinson D, Brierley J. Making decisions to limit treatment in life-limiting and life-threatening conditions in children: a framework for practice. Arch Dis Child 2015;100(Suppl 2):s1-s23.

300. Ray S, Brierley J, Bush A, et al. Towards developing an ethical framework for decision making in long-term ventilation in children. Arch Dis Child. 2018;103(11):1080-4.

301. Brierley J, Larcher V. Organ donation from children: Time for legal, ethical and cultural change. Acta Paediatrica. 2011;100(9):1175-9.

302. Rabiee P, Sloper P, Beresford B. Doing research with children and young people who do not use speech for communication. Child Soc. 2005;19(5):385-96.

303. Carroll C, Sixsmith J. Exploring the facilitation of young children with disabilities in research about their early intervention service. Child Lang Teach Therapy. 2016;32(3):313-25.

304. Hudson BF, Oostendorp L, Candy B, et al. The under reporting of recruitment strategies in research with children with life-threatening illnesses: A systematic review. Palliat Med. 2017;31(5):419-36.

305. Passamano L, Taglia A, Palladino A, et al. Improvement of survival in Duchenne Muscular Dystrophy: retrospective analysis of 835 patients. Acta Myol. 2012;31(2):121-5.

306. Hurley M, McKeever T, Prayle A, et al. Rate of improvement of CF life expectancy exceeds that of general population--observational death registration study. J Cyst Fibros. 2014;13(4):410-5.

307. Saunders B, Sim J, Kingstone T, et al. Saturation in qualitative research: exploring its conceptualization and operationalization. Qual Quant. 2018;52(4):1893-907.

308. Lanham H, Leykum L, Taylor B, et al. How complexity science can inform scale-up and spread in health care: understanding the role of self-organization in variation across local contexts. Soc Sci Med. 2013;93:194-202.

309. Twamley K, Kelly P, Moss R, Mancini A, Craig F, Koh M, et al. Palliative care education in neonatal units: impact on knowledge and attitudes. BMJ Support Palliat Care. 2013;3(2):213-20.

310. Witkamp F, Heide A, Der Rijt C, et al. Effect of a Palliative Care Nurse Champion Program on Nursing Care of Dying Patients in the Hospital: A Controlled before and after Study. Palliat Med. 2016. 30(2):180-8. doi: 10.1177/0269216315588008

311. Manson J, Blomfield H, Patterson L, et al. 19 Do palliative care champions increase staff confidence to have difficult conversations? 2019. BMJ Support Palliat Care;9(Suppl 1):A7-A.

312. Widger K, Seow H, Rapoport A, et al. Children's End-of-Life Health Care Use and Cost. Pediatrics. 2017; 139 (4) DOI: 10.1542/peds.2016-2956.

313. Feudtner C, Kang T, Hexem K, et al. Pediatric palliative care patients: a prospective multicenter cohort study. Pediatrics. 2011;127(6):1094-101.

314. Booth A, Maddison J, Wright, K et al. Research prioritisation exercises related to the care of children and young people with life-limiting conditions, their parents and all those who care for them: A systematic scoping review. Palliat Med. 2018;32(10):1552-66.

Appendix 1: Publications from the PhD

Specialist paediatric palliative care services: what are the benefits?

Sarah Mitchell,[1] Andrew Morris,[1] Karina Bennett,[1] Laiba Sajid,[2] Jeremy Dale[1]

[1]Warwick Medical School, University of Warwick, Coventry, UK
[2]Sibling Council, Acorns Children's Hospice Trust, Birmingham, UK

Correspondence to
Dr Sarah Mitchell, Warwick Medical School, University of Warwick, Gibbet Hill Road, Coventry CV4 7AL, UK;

ABSTRACT

Background The number of children and young people (CYP) living with life-limiting and life-threatening conditions is rising. Paediatric palliative care is a relatively new aspect of healthcare, the delivery of which is variable, with a wide range of healthcare and voluntary sector providers involved. Policy recommendations are for Specialist Paediatric Palliative Care (SPPC) services to be supported by a physician with specialist training.

Aim To examine the research evidence regarding the distinct benefits of SPPC services, with 'Specialist Paediatric Palliative Care' defined as palliative care services supported by a specialist physician.

Method Systematic review of studies of SPPC services published in English from 1980 to 2016. Keyword searches were carried out in medical databases (Cochrane, PubMed, EMBASE, CINAHL and AMED) and a narrative synthesis.

Results Eight studies were identified, most of which were retrospective surveys undertaken within single institutions; three were surveys of bereaved parents and three were medical notes reviews. Together they represented a heterogeneous body of low-level evidence. Cross-cutting themes suggest that SPPC services improve the quality of life and symptom control and can impact positively on place of care and family support.

Conclusions Current evidence indicates that SPPC services contribute beneficially to the care and experience of CYP and their families, but is limited in terms of quantity, methodological rigour and generalisability. Further research is necessary given the significant workforce and resource implications associated with policy recommendations about the future provision of SPPC and to address the need for evidence to inform the design and delivery of SPPC services.

> ### What is already known on this topic?
>
> ► The number of children and young people (CYP) living with life-limiting and life-threatening conditions is rising with continuing advances in clinical medicine.
> ► There are international recommendations and standards for Specialist Paediatric Palliative Care (SPPC) services, but this is a relatively new subspecialty and is inconsistently available.
> ► The more universal adoption of recommendations and standards requires significant investment of resource, which is difficult to achieve.

> ### What this study adds?
>
> ► This is the first systematic review of research related to the evaluation of SPPC.
> ► The review identifies a summary of the evidence that suggests that SPPC provides benefit to CYP and families.
> ► Key themes have been identified to inform future service development and research in paediatric palliative care.

INTRODUCTION

With advances in medical treatments and the use of medical technology, a growing number of children and young people (CYP) live with life-limiting conditions (LLCs) and life-threatening conditions (LTCs).[1 2] This includes CYP who live with conditions where curative treatment is feasible but can fail, while for others, there is no known cure.[3] Ensuring sustainable healthcare services that can effectively meet the needs of these CYP and those of their families presents a significant challenge.

Palliative care for CYP has been defined as "an active and total approach to care, from the point of diagnosis or recognition, embracing physical, emotional, social and spiritual elements through to death and beyond. It focuses on enhancement of the quality of life for the CYP and support for the family and includes the management of distressing symptoms, provision of short breaks and care through death and bereavement.[3]" Globally, it is estimated that seven million CYP (aged 0–19) could benefit from palliative care services,[4] with at least 49 000 in the UK.[5] However, most countries have no paediatric palliative care services, including hospices.[4] Where paediatric palliative care services have developed, this has been largely as a result of the determination of motivated individuals and charitable funders.[6 7] The type and availability of services vary geographically due to the wide range of healthcare and voluntary sector providers involved.[4]

Specialist Paediatric Palliative Care (SPPC) services are defined in UK and European standards as those supported by a physician with specialty training (a consultant) in paediatric palliative medicine.[1 8–10] However, SPPC is yet to become an established medical subspecialty, and few countries have doctors trained to this level.[4] As a result, there is a tension between this standard of care and the many existing services which specialise in the

provision of paediatric palliative care but lack the support of specialty-trained physicians.

OBJECTIVE

The objective of this systematic review is to contribute to the debate regarding the design of paediatric palliative care services by specifically examining SPPC, defined as a palliative care service supported by a physician with specialty training in paediatric palliative medicine, and asking "What are the distinct benefits of these SPPC services to CYP and their families?" The review also provides an opportunity to identify evidence gaps for further research.

DESIGN

The Centre for Reviews and Dissemination (CRD) guidance for systematic reviews in healthcare and the Cochrane Collaboration's Handbook for Systematic Reviews of Interventions informed the review's methodology.[11][12] The structure and content were informed by the preferred reporting items for systematic reviews and meta-analyses (PRISMA) guidelines.[13] A protocol has been registered and published on the PROSPERO database (ref no: CRD42016050677).

Search strategy
Information sources
The following electronic databases were searched from September 2015 to January 2016 with the last search on 7 January 2016:
- Cochrane Central Register of Controlled Trials and Cochrane Database of Systematic Reviews
- PubMed (1980 onwards)
- EMBASE (1980 onwards)
- CINAHL (1981 onwards)
- AMED (1985 onwards)

After initial broad scoping searches, the search terms outlined in table 1 were used to perform a focused systematic search. The population search was carried out first, followed by the intervention search. The search was carried out with the advice of the University of Warwick specialist librarian. Hand searching of references, 'cited by' and PubMed-related articles link searches were also carried out.

Inclusion/exclusion criteria
Inclusion and exclusion criteria are outlined in table 2.

Study selection
Duplicate articles were removed. Titles and abstracts were screened, followed by examination of the full text. Articles were assessed for inclusion independently by three reviewers (SM, KB and AM).

Data management
Two reviewers extracted relevant data to an Excel spreadsheet (AM and KB), which was independently checked for accuracy and detail by SM. The team discussed any disagreements.

Data synthesis
The included studies were compared and contrasted using a data extraction table. There were no comparable statistics and therefore a systematic narrative synthesis[14] was undertaken, identifying cross-cutting themes from each study. The narrative was reviewed at intervals by LS, our patient and public involvement coauthor, with feedback provided on relevance to family experience and by JD for intellectual content.

RESULTS
Study selection
Seven hundred and seventy relevant articles were identified. Seven hundred and fifty-five were excluded after title and abstract screening and the removal of duplicates, leaving 15 articles. Three of these were conference abstracts of ongoing studies which were not available as full-text articles or as unpublished studies from the authors and were therefore excluded. After applying the inclusion and exclusion criteria to the remaining 12 articles, 4 were excluded because they did not concern SPPC services with specialist medical support, leaving 8 articles. This process is shown in figure 1. Study characteristics are summarised in table 3.

Study location
Included studies were from the USA,[15–18] the UK,[19] Germany,[20] Australia[21] and Canada.[22]

Study quality
The studies represented a heterogeneous body of evidence;[13] seven were retrospective studies;[15–19 21 22] three were surveys of bereaved parents,[15 17 22] one was an epidemiological study[19] and three were medical notes reviews,[16 18 21] one of which included an economic analysis.[18] There was one prospective longitudinal survey.[20] There were no randomised-controlled trials or systematic reviews. All had clear aims and used appropriate methodology and approached the ethical issues. All acknowledged the limitations in their study design and recruitment strategies, and data were collected in a way that would address the research aims. All gave clear descriptions of their data analysis, results and findings.[23]

All had been published since 2012 and were carried out within single institutions or services. The largest study in terms of patient numbers was an epidemiological study, which looked at data regarding 2508 CYP but was limited by missing data items.[19] The notes review studies examined the care of a total of 611 CYP.[16 18 21] Three studies concerned only CYP with cancer.[15 19 22] The other five studies concerned services for CYP with non-malignant conditions as well as those with cancer.[16–18 20 21]

Four studies made use of questionnaires with caregivers;[15 17 20 22] three were surveys of bereaved parents.[15 17 22] The total number of bereaved parents included in these studies was 200. Time since bereavement ranged from 7 months to over 4 years.[15 17 22] Response rates for postal surveys of a total number of bereaved parents in a time period were 65/192 (37%)[17] and 60/166 (36%).[15] A response rate of 75/140 (75%)
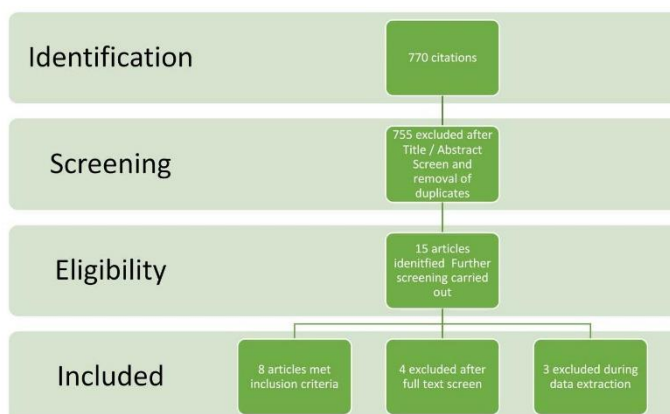
Table 1	Search strategy
Population	Pediatr*/Paediatr* AND (Infant OR Child* OR Adolescen*)
Intervention	Palliat* Special* End-of-life Hospice Terminal care Consultant Physician Delivery of healthcare Service

Table 2: Inclusion and exclusion criteria

PICOS dimension	Inclusion	Exclusion
Population	Children and young people aged from 0 to 18 years (inclusive)	Studies concerning neonatal palliative care Studies concerning specifically young people making the transition to adult services Adult studies
Intervention	'Specialist Paediatric Palliative Care' defined as a palliative care service supported by a physician with specialty training in paediatric palliative medicine, as per current recommendations and service specifications	Paediatric palliative care services that did not meet the specialist specification, including hospice services, and services supported by paediatricians who had not received specialty training in paediatric palliative medicine (where it was possible to establish this) Neonatal palliative care services Adult palliative care services (who may be catering for paediatric patients) Any other usual care
Comparator	Usual care or palliative care that was provided by other types of services	
Outcomes	Any formal measure of evaluation concerning the acceptability or effectiveness of the intervention	
Study design	Any evaluative study design	Review articles, descriptive or theoretical papers that did not present original research findings
Publication	Databases were searched from 1980 onwards Unpublished grey literature	Non-English language papers Articles only available in abstract form where no full text is available (the authors were contacted) Voluntary sector reports

PICOS, Population, Intervention, Comparators, Outcome, Study design.

Figure 1 Preferred reporting items for systematic reviews and meta-analyses (PRISMA) flow diagram.

Identification — 770 citations

Screening — 755 excluded after Title / Abstract Screen and removal of duplicates

Eligibility — 15 articles idenitfied Further screening carried out

Included — 8 articles met inclusion criteria | 4 excluded after full text screen | 3 excluded during data extraction

was achieved where eligibility criteria were applied.[22] The highest response rate for a questionnaire survey was 93% (40/43), with the questionnaire administered face to face with family members at the time they were receiving care from the SPPC services.[20] This study also attempted the assessment of children by self-report but due to young age and clinical condition this was possible with only three CYP.[20]

Key themes

Four key themes about how SPPC services can impact on CYP and their families were identified. These related to:
1. Quality of life
2. Symptom control
3. Place of care
4. Family support

Quality of life

The studies provide evidence that SPPC services contribute to improving the quality of life of CYP and family through emotional support, care planning and help with medical decision making,[16 17 20 21] as well as through the management of distressing physical symptoms.[17 21] Improved satisfaction with care was reported once SPPC services were involved.[16 20] One study reported that CYP who were in contact with a SPPC "had more fun and [were] more likely to have an experience which added meaning to their life"[15] than those who were not.

Symptom control

Pain and symptom management was described as one of the main reasons for referral to SPPC.[21] Improvements in children's symptom control with the involvement of the SPPC team were reported by parents retrospectively.[15 20] With the involvement of SPPC services, more care, including symptom management, was delivered in the home environment,[15] alongside other support for caregivers and practical support.[20]

Place of care

Three studies provided evidence to suggest that referral to SPPC is associated with fewer admissions to hospital[19 20] or a reduced length of stay.[18] The involvement of SPPC services was associated

3

Table 3 Study characteristics

Study	Design and research questions	Participants	Intervention	Findings
Sheetz et al[17]	What are parents' perceptions about whether a SPPC programme was providing key elements of paediatric palliative care? Are parents satisfied with the service? Questionnaire survey of parents whose children were receiving care from a specific SPPC programme. Salt Lake City, USA.	Parents of 65 children who had died while under the care of the programme. CYP with a range of LLCs and LTCs, including cancer and complex chronic conditions, most frequently genetic/congenital, neuromuscular and cardiovascular conditions.	SPPC programme: multi-disciplinary team including paediatrician and nurse practitioner board-certified in paediatric palliative medicine, nurse manager, social worker, chaplain. Hospital-based inpatient consultation service and home consultation service after discharge. Age range prenatal–36 years.	65/192 responded (37%). 37%[77] of children had died at home, 53%[17] at the hospital, 8%[5] at another hospital and 2%[1] in a long-term care facility. 21 were also receiving hospice services at the time of death. 95% agreed or strongly agreed that the SPPC team helped them make decisions about their child's care, 5% disagreed. 76% felt the team had helped them set goals for care (others unsure or disagreed), 78% agreed that those goals of care were subsequently met. 13% unsure, 10% disagreed. 95% felt supported in their choices for their child's care. Also, 78% felt that physical pain/distressing symptoms were controlled at the end of life, 22% unsure or disagreed. 71% felt symptoms other than pain were controlled adequately. High levels of parent satisfaction with SPPC. SPPC had an important role in education: decision making, the process of death and aspects of the medical system. Feedback included a desire that the team were involved sooner.
Fraser et al[19]	What is the impact of Specialist Paediatric Palliative Care Service (SPPCS) on the number of hospital admissions in children before their death with cancer? Retrospective cohort study of epidemiological data. Differences in hospital admission patterns were assessed using negative binomial regression. Yorkshire, UK.	2508 patients aged 0–19 years with cancer from 1990 to 2009, who were in the catchment area for the SPPCS.	SPPC based at a children's (aged up to 35 years) hospice with a full-time consultant from 2004. 24/7 on-call medical service and a home visiting service.	27.7% of those who had died were referred to SPPC (less than a third). 182 had died and had been referred, 475 had not been referred before they died. No significant difference in terms of demographics. Most commonly referred were those with central nervous system (CNS) tumours. Referral to SPPC was associated with a significantly lower rate of planned hospital admissions (incidence rate ratio=0.60, 95% CI 0.43 to 0.85). There was no significant difference in emergency (which comprised 97% of admissions) or total hospital admission rates. Children with CNS tumours had significantly less hospital admissions compared with those with leukaemia (may be due to the nature of treatment).
Groh et al[20]	Does the involvement of a SPPC team (home care) address the needs of patients and their families and thus lead to an increase in acceptance and effectiveness of SPPC? Prospective non-randomised study with two validated questionnaires; the first during the first week of paediatric palliative home care (SPPC) service involvement, and the second a few days—6 months later depending on the clinical condition. Face to face by a trained psychologist. Bavaria, Germany.	All primary caregivers of severely ill children receiving SPPC via the Paediatric Palliative Home Care (PPHC) team for the first time between April 2011 and June 2012. 40 families. 18 CYP died before study ended. 16 of them at home. Wide range of diagnoses, including cancer and complex chronic conditions.	Multiprofessional SPPC team consisting of three paediatricians, two nurses, a social worker and a chaplain, all with special training in palliative care. 24/7 on call.	SPPC was associated with improved satisfaction with care and quality of care. Significant improvement in children's symptom control and quality of life as perceived by parents. Parents own quality of life and burden relief significantly increased and psychological distress and burden decreased. SPPC led to reduced rates of hospitalisation and improved caregiver satisfaction with care received, including psychological support and support of activities of daily living. Caregivers felt more informed on disease situation and progression, better taken care of and improved communication with child. Psychosocial support was identified as the most helpful aspect of care. 24/7 on-call service and time for detailed conversations highly valued, as were support with practicalities, for example, Advance Care Planning, equipment.
Niswander et al[16]	What SPPC are CYP receiving at home, and how is their end-of-life experience perceived by parents and medical personnel? Retrospective cohort review of medical notes of children who died from December 2004 to May 2008. New York, USA.	36 children who died with a wide range of diagnoses, including cancer, enrolled in the programme for a median 1.1 years before they died.	A team consisting of paediatric nurses, paediatric nurse practitioners, a paediatrician board-certified in hospice and palliative medicine, child life specialists, social workers, chaplain and expressive therapists.	There was a median of three hospitalisations (including planned), two emergency room visits in the last 6 months of life. Median of 24 home visits (1–121), home visit frequency varied. Symptom control was important, as were 'goals of care' discussions, which happened a median of 16 days before death (0–116). 25 had home Do Not Attempt Cardiopulmonary Resuscitation (DNACPR). 15 CYP were aware of impending death (recorded)—tended to be older CYP. 16 children died at home, 20 died in hospital. 16 had recorded preferred place of death. Conclusion was that children who die of complex chronic conditions spend most of their last 6 months at home; community SPPC contributes substantially to their care and comfort.

Table 3 Continued

Study	Design and research questions	Participants	Intervention	Findings
Postier et al[18]	What is the healthcare utilisation by children prior to enrolment in SPPC compared with the period after enrolment? Retrospective cohort study of electronic medical records and economic analysis. Minneapolis, USA.	425 children with a range of diagnoses aged 1–21 under the care of SPPC for at least 1-day between 2000 and 2010.	24/7 access and care coordination through home visits by nurses, social workers, child life specialists, chaplains, music/massage therapists, physicians and volunteers.	No. of hospital admissions didn't change. Length of stay decreased by 2 weeks post-SPPC involvement, with a significant drop in healthcare costs. Largest decreases in resource utilisation for the non-malignant group—reduced length of stay in hospital with SPPC involvement, decrease in hospital charges for those under SPPC >6 months.
Herbert et al[21]	What are the characteristics of the population cared for by the SPPC service? What outcomes are the SPPC achieving and how has the service developed? Retrospective cohort review of medical notes of children who were referred to the service in a 2-year period. Brisbane, Australia.	150 patients refered over a 24-month period. Wide range of diagnoses.	The SPPC developed from the existing paediatric oncology palliative care service at the Royal Children's Hospital (RCH) and incrementally expanded over a period of 3.5 years, commencing with a dedicated paediatrician trained in palliative care, followed by nursing staff and the addition of dedicated allied health staff from 2010. Offers biannual training days and videoconferencing. 24/7 telephone support.	Median duration of contact time with the service was 83 days. Non-oncology diagnosis was likely to result in longer use of the service (>6 months). 41% of children died at home and 48% died at hospital. Reasons for referral: pain and symptom management (29%), advanced care planning (25%), community care planning (21%) and end-of-life care (26%).
Friedrichsdorf et al[15]	How does end-of-life pain and symptom management in children with advanced cancer who received care exclusively from oncology compare with those who received concurrent SPPC home care? What are the differences between the two groups regarding health outcomes, including quality of life and location of death. Retrospective cohort survey study of bereaved parents whose children died of cancer. Minneapolis, USA.	Final sample of 60 bereaved parents of children who died of cancer between 2002 and 2008 at a US tertiary paediatric institution.	Paediatric palliative care (PPC) PPC nurses, social workers and chaplaincy. A PPC physician and/or paediatric oncologist or oncology advanced practice registered nurse. 24/7 nursing on-call for home visits.	No significant difference in prevalence of symptoms between those referred to SPPC and those who were not. There was a trend towards greater perceived suffering from pain in the group who were not referred to SPPC group. Seizures and nausea/vomiting were the most successfully managed in both groups. More parents in SPPC group wanted their child to die at home and had an opportunity to plan this. More children in PPC group did die at home. SPPC children had more fun and more likely to have an experience, which added meaning to their life.
Kassam et al[22]	Is referral to SPPC associated with improved end-of-life care communication for children with advanced cancer and their families? Questionnaire survey and medical record review examining differences in end-of-life communication for children with advanced cancer who were referred to a SPPC team. Toronto, Canada.	75 bereaved parents	Paediatric Advanced Care Team (PACT) team: four palliative care physicians, three nurses, two grief support coordinators and one administrative assistant. PACT also draws on the special knowledge of other professionals and volunteers like chaplains, social workers, nurses, bioethicists, physicians, pain experts, volunteers and parents.	Most frequent communication was DNACPR discussion. Least frequent was discussion of death and dying when appropriate and advice on how to talk to child about this. Univariate analysis showed parents more likely to have the following five communication elements if a palliative care team were involved: Discussion of death and dying with parents and with the child, guidance on how to talk to their child, preparing parents for medical aspects of death and sibling support.

CYP, children and young people; LLCs, life-limiting conditions; LTCs, life-threatening conditions; SPPC, Specialist Paediatric Palliative Care.

with care planning discussions and considering a preferred location of death.[16 22] More CYP died at home with SPPC involvement than not.[15 20] Differences in terms of both diagnosis and geographical location of the family home contributed to the location of death. In one study, CYP with a cancer diagnosis were more likely to die at home if they lived in a rural location; CYP with non-malignant disease were more likely to die in a tertiary hospital.[21] However, there was evidence that 'goals of care' discussions tended to occur relatively late, with the median time before death that this discussion took place being 16 days.[16]

Family experience

There were several areas in which SPPC intervention was reported as contributing to an improvement in family members' quality of life.[20 22] Access to services 24 hours a day, 7 days/week was valued.[20] Perception of psychological support and support for carrying out 'day-to-day activities' increased, and there was a decrease in anxiety and depression among parents.[20] SPPC teams provided support with medical decision making,[16 17] including discussions about resuscitation,[16 22] help with communication between family members, including with their child, and with other healthcare teams.[17 20 22] The SPPC team played an important role in educating parents about both the process of death and aspects of the medical system.[17]

Low referral rates to SPPC services were described[19] and the average length of time that a child was under the care of SPPC varied from 20 days to over 1 year.[16 18 21 22] Feedback from families included a desire that SPPC was involved earlier on in the course of a child's illness.[17]

DISCUSSION

This review set out to investigate the current evidence regarding the distinct benefits to CYP and their families of care provided by SPPC, defined as a palliative care service supported by a physician with specialist training.

A small number of studies met the inclusion criteria, all of which had been published within the last 5 years. All of these were single-centre studies with relatively small patient numbers and rank low in the hierarchy of evidence due to their methodological limitations.[12] This is a well-recognised problem in palliative care research, due to clinical and ethical challenges, and brings into question the value of systematic reviews in this subject area.[24] Specific methodological challenges exist around gatekeeping of participants by clinicians,[25] which was described as a barrier to recruitment in one study.[22] In surveys, families were found to be unreachable by phone or post, did not respond or were ineligible to participate due to language barriers.[15 17 20 22] Where families did respond, they are likely to have been particularly motivated to participate, and therefore the survey findings may not be generalisable to a more diverse population of families. One study tried to collect the views of CYP, but found this was not possible.[20]

Benefits of SPPC

Despite the limited quality of the evidence, there are cross-cutting themes from the eight studies suggesting that SPPC may enable improved quality of life for CYP and parents, improved symptom control, has an impact on the place of care and an increased likelihood that a preferred place of death is achieved.[16 17 20–22]

Service delivery

The review is timely as the specialty of paediatric palliative medicine further develops, with international standards,

specifications and recommendations.[8–10 26] Systematic consideration of the available evidence to support the development of services and policy is necessary, particularly as the number of CYP with LLCs and LTCs rises.

There is ongoing emphasis on place of death as an outcome measure in palliative care, despite a limited evidence base to support this.[27] 'Choice' in end-of-life care is frequently highlighted in policy,[28] and families desire the option of care being provided at home.[29] This review suggests an association between referral to a SPPC team and opportunity to first express preferences for goals of care and location of death and then achieve it,[15 16 22] although there was some evidence that this occurred relatively late in the course of illness.[16] Key factors that enable these discussions have been described as continuous relationships, time for open, honest conversations and the provision of symptom control.[15 20 29 30]

Although adequate control of symptoms was not always perceived by parents,[15 17] there was evidence to suggest that more effective symptom control could be delivered in the home environment with the involvement of an SPPC team.[15] Further research into symptom management in CYP, including the use of medications and routes of administration, and how this can be delivered in both community and inpatient care settings, is an important focus for SPPC.

SPPC service design

What cannot be ascertained from the available evidence is which elements of SPPC services are associated with the benefits described, the mechanisms by which these benefits occur or the impact of the presence of a specialty-trained physician. This review looked specifically at services with a specialty-trained physician and excluded studies of any other model of care. However, nurse-led paediatric palliative care services and children's hospice services have also been shown to benefit CYP and their families, particularly in terms of place of care,[31–34] coordination of care[35] and family support.[36] Research to compare more clearly the different types of services, and how they can effectively work together, would be valuable. Further research to investigate the most effective services for children with differing LLCs would also be of value given the wide variation in disease trajectories, family need and outcomes.[3]

The benefits of a specialist physician in a service have been broadly described as advanced clinical expertise and academic, educational and strategic leadership,[37] all of which are important in SPPC as the specialty develops. Securing funding to develop both specialty training and new consultant posts presents a major challenge and will require clear business cases. Future innovation and development of the SPPC workforce and the implementation of new policies, including National Institute for Health and Care Excellence (NICE) guidance,[26] should be accompanied by robust plans for evaluation.

This review identified only one study which made reference to the value of parental input into the development of SPPC services.[17] Codesign of services with CYP and families[28 38] and work to address possible reasons for low referral rates to SPPC, such as negative perceptions of palliative care among families[39] and healthcare professionals,[40 41] would be highly relevant.

CONCLUSION

Future service development recommendations should address the need for accessible and sustainable SPPC services for all CYP who need them. However, as this review demonstrates, there is limited evidence to inform policy guidance within the overall provision of paediatric palliative care. In the context of a

growing number of CYP and families who could benefit from SPPC, there is a need for further research, innovation and debate. Robust evaluation of services, care models and professional roles, as well as research to understand the mechanisms by which benefits are delivered to CYP and families, is necessary. These are key considerations for those who are leading the development of SPPC and for service commissioners.

REFERENCES

1 Hain R, Heckford E, McCulloch R. Paediatric palliative medicine in the UK: past, present, future. *Arch Dis Child* 2012;97:381–4.
2 Harrop E, Edwards C. How and when to refer a child for specialist paediatric palliative care. *Arch of Dis Child Ed Pract* 2013;98:202–8.
3 Together for Short Lives. Children's Palliative Care Definitions. http://www.togetherforshortlives.org.uk/assets/0000/4089/CPC_definitions.pdf
4 Knapp C, Woodworth L, Wright M, *et al*. Pediatric palliative care provision around the world: a systematic review. *Pediatr Blood Canc* 2011;57:361–8.
5 Fraser LK, Miller M, Hain R, *et al*. Rising national prevalence of life-limiting conditions in children in England. *Pediatrics* 2012;129:e923–9.
6 MacNamara-Goodger K, Feudtner C. History and epidemiology. In: Goldman A, Hain R, Liben S, eds. *Oxford textbook of palliative care for children*. 2nd edn. Oxford: Oxford University Press, 2012:3–4.
7 Beringer AJ, Eaton NM, Jones GL. Providing a children's palliative care service in the community through fixed-term grants: the staff perspective. *Child Care Health Dev* 2007;33:619–24.
8 European Association for Palliative Care TSG. IMPaCCT: standards for paediatric palliative care in Europe. *Eur J Palliat Care* 2007;14:109–14.
9 *NHPCO's pediatric standards: a key step in advancing care for America's children [press release]*. National Hospice and Palliative Care Organization, 2009. http://www.nhpco.org/quality/nhpco%E2%80%99s-standards-pediatric-care
10 NHS England. *E03/S/h—2013/14 NHS standard contract for paediatric medicine: palliative care particulars, schedule 2—the services, A—service specifications*. NHS England, 2013. https://www.england.nhs.uk/wp-content/uploads/2013/06/e03-paedi-med-pall.pdf
11 Centre for Reviews and Dissemination. *Systematic Reviews: CRD's guidance for undertaking reviews in health care*. University of York, 2008.
12 Higgins P, Green S, eds. *Cochrane Handbook for Systematic Reviews of Interventions*. UK: Wiley-Blackwell and The Cochrane Collaboration, 2011.
13 Shamseer L, Moher D, Clarke M, *et al*. Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P) 2015: elaboration and explanation. *Br Med J* 2015;349:g7647.
14 Popay J, Roberts H, Sowden A, *et al*. Guidance on the conduct of narrative synthesis in systematic reviews. Lancaster: ESRC Research Methods Programme, 2006.
15 Friedrichsdorf SJ, Postier A, Dreyfus J, *et al*. Improved quality of life at end of life related to home-based palliative care in children with cancer. *J Palliat Med* 2015;18:143–50.
16 Niswander LM, Cromwell P, Chirico J, *et al*. End-of-life care for children enrolled in a community-based pediatric palliative care program. *J Palliat Med* 2014;17:589–91.
17 Sheetz MJ, Bowman MAS. Parents' perceptions of a pediatric palliative program. *Am J Hosp Palliat Med* 2013;30:291–6.
18 Postier A, Chrastek J, Nugent S, *et al*. Exposure to home-based pediatric palliative and hospice care and its impact on hospital and emergency care charges at a single institution. *J Palliat Med* 2014;17:183–8.
19 Fraser LK, van Laar M, Miller M, *et al*. Does referral to specialist paediatric palliative care services reduce hospital admissions in oncology patients at the end of life? *Br J Canc* 2013;108:1273–9.
20 Groh G, Borasio GD, Nickolay C, *et al*. Specialized pediatric palliative home care: a prospective evaluation. *J Palliat Med* 2013;16:1588–94.
21 Herbert A, Bradford N, Donovan L, *et al*. Development of a state-wide pediatric palliative care service in Australia: referral and outcomes over two years. *J Palliat Med* 2014;17:288–95.
22 Kassam A, Skiadaresis J, Alexander S, *et al*. Differences in end-of-life communication for children with advanced cancer who were referred to a palliative care team. *Pediatr Blood Canc* 2015;62:1409–13.
23 http://www.casp-uk.net/casp-tools-checklists
24 Wee B, Hadley G, Derry S. How useful are systematic reviews for informing care practice? Survey of 25 Cochrane systematic reviews. *BMC Palliat Care* 2008;7:13.
25 Crocker JC, Beecham E, Kelly P, *et al*. Inviting parents to take part in paediatric palliative care research: a mixed-methods examination of selection bias. *Palliat Med* 2015;29:231–40.
26 National Institute for Health and Care Excellence (NICE). *NICE Guideline NG61: End of Life Care for Infants, Children and Young People*. 2016. https://www.nice.org.uk/guidance/ng61
27 Bluebond-Langner M, Beecham E, Candy B, *et al*. Preferred place of death for children and young people with life-limiting and life-threatening conditions: a systematic review of the literature and recommendations for future inquiry and policy. *Palliat Med* 2013;27:705–13.
28 NHS England. Delivering the Forward View: NHS planning guidance 2016/17–2020/21. 2015. https://www.england.nhs.uk/wp-content/uploads/2015/12/planning-guid-16-17-20-21.pdf
29 Coad J, Kaur J, Ashley N, *et al*. Exploring the perceived met and unmet need of life-limited children, young people and families. *J Pediatr Nurs* 2015;30:45–53.
30 Wolfe J, Grier HE, Klar N, *et al*. Symptoms and suffering at the end of life in children with cancer. *N Engl J Med* 2000;342:326–33.
31 Breen M, Anderson A-K, Jones KP. Evaluation of place of care at time of death at one United Kingdom principal treatment centre for children with cancer. *Europ J Onc Nurs* 2011;15:277.
32 Sastry J, Ronghe M, Murphy D, *et al*. Nurse led palliative care at home for children with brain tumours-west of Scotland experience. *Neuro-Onc* 2014;16:i109.
33 Mastro KA, Johnson JE, McElvery N, *et al*. The benefits of a nurse-driven patient-and family-centred pediatric palliative care program. *J Nurs Admin* 2015;45:423–8.
34 Martinson IM, Armstrong GD, Geis DP, *et al*. Home care for children dying of cancer. *Pediatr* 1978;62:106–13.
35 Junger S, Hasan C, Burger A, *et al*. Coordination of paediatric palliative home care services. *Palliat Med* 2010;1:S22.
36 Van Der Tempel A, Groenenberg AP, Vallianatos S. Case management in paediatric palliative care. *Arch Dis Child* 2014;99:A536–7.
37 Academy of Medical Royal Colleges. *The benefits of consultant-delivered care*. London: Academy of Medical Royal Colleges, 2012.
38 National Palliative and End of Life Care Partnership. Ambitions for Palliative and End of Life Care: A national framework for local action. 2015. http://endoflifecareambitions.org.uk/wp-content/uploads/2015/09/Ambitions-for-Palliative-and-End-of-Life-Care.pdf
39 Morstad Boldt A, Yusuf F, Himelstein BP. Perceptions of the term palliative care. *J Palliat Med* 2006;9:1128–36.
40 Twamley K, Craig F, Kelly P, *et al*. Underlying barriers to referral to paediatric palliative care services: knowledge and attitudes of health care professionals in a paediatric tertiary care centre in the United Kingdom. *J Child Health Care* 2014;18:19–30.
41 Monterosso L, Kristjanson LJ. Supportive and palliative care needs of families of children who die from cancer: an Australian study. *Palliat Med* 2008;22:59–69.

Specialist paediatric palliative care services: what are the benefits?

Sarah Mitchell, Andrew Morris, Karina Bennett, Laiba Sajid and Jeremy Dale

	Updated information and services can be found at: http://adc.bmj.com/content/early/2017/04/04/archdischild-2016-31202 6

These include:

References	This article cites 29 articles, 7 of which you can access for free at: http://adc.bmj.com/content/early/2017/04/04/archdischild-2016-31202 6#BIBL
Email alerting service	Receive free email alerts when new articles cite this article. Sign up in the box at the top right corner of the online article.

Notes

BMJ Open

The journey through care: study protocol for a longitudinal qualitative interview study to investigate the healthcare experiences and preferences of children and young people with life-limiting and life-threatening conditions and their families in the West Midlands, UK

Sarah Mitchell,[1] Anne-Marie Slowther,[1] Jane Coad,[2] Jeremy Dale[1]

Check for updates

¹Warwick Medical School, University of Warwick, Coventry, UK
²Faculty of Health and Life Sciences, Coventry University, Coventry, UK

Correspondence to
Dr Sarah Mitchell;

ABSTRACT

Introduction The number of children and young people living with life-limiting and life-threatening conditions is rising. Providing high-quality, responsive healthcare for them and for their families presents a significant challenge. Their conditions are often complex and highly unpredictable. Palliative care is advocated for people with life-limiting and life-threatening conditions, but these services for children are highly variable in terms of availability and scope. Little is known about the lived experiences and preferences of children and their families in terms of the palliative care that they do, or do not, receive. This study aims to produce an in-depth insight into the experiences and preferences of such children and families in order to develop recommendations for the future provision of services. The study will be carried out in the West Midlands, UK.

Methods and analysis A qualitative study comprising longitudinal interviews over a 12-month period with children (aged 5–18 years) living with life-limiting or life-threatening conditions and their family members. Data analysis will start with thematic analysis, followed by narrative and cross-case analysis to examine changing experiences and preferences over time, at the family level and within the wider healthcare system. Patient and public involvement (PPI) has informed the design and conduct of the study. Findings will be used to develop recommendations for an integrated model of palliative care for children in partnership with the patient and public involvement (PPI) group.

Ethics and dissemination Ethical approval was granted in September 2016 by the National Health Service Health Research Authority (IRAS ID: 196816, REC reference: 16/WM/0272). Findings will be of immediate relevance to healthcare providers, policy-makers, commissioners and voluntary sector organisations in the UK and internationally. Reports will be prepared for these audiences, as well as for children and their families, alongside academic outputs.

Strengths and limitations of this study

▶ An in-depth, contextual, longitudinal qualitative study with multiple child and family member stories captured over time.

▶ New insights will be provided because all of the children and families included in the study could benefit from palliative care as it is currently defined, however, not all will have had conversations about this or have been referred to specialist palliative care services. Findings will focus on healthcare, but there is a wider applicability and relevance to social care and joint planning of services.

▶ A diverse study population in terms of age, clinical condition, cultural background and family structure will allow detailed consideration of the role of healthcare services in effectively recognising and supporting children and families with their individual needs. However, all will speak English.

▶ Neonates, preschool children and young people at transition (over the age of 18 years) are all excluded and warrant research in their own right.

▶ There are multiple potential sources of bias which will be addressed throughout the study, including recruitment bias and the unconscious bias of the researcher.

INTRODUCTION

Children and young people with life-limiting conditions and life-threatening conditions represent a growing concern in healthcare.[1] With advances in clinical practice, the number of children living with these conditions is rising.[1-3] The nature of their conditions is complex and unpredictable; the risk of a sudden deterioration and death is an

---

Box   Children's palliative care definitions[15 62]

Palliative care for children with life-threatening conditions is defined by WHO as 'a special, albeit closely related field to adult palliative care; the principles apply to other paediatric chronic disorders:

► The active total care of the child's body, mind and spirit, and support to the family.
► It begins when illness is diagnosed, and continues regardless of whether or not a child receives treatment directed at the disease.
► Health providers must evaluate and alleviate a child's physical, psychological and social distress.
► Effective palliative care requires a broad multidisciplinary approach that includes the family and makes use of available community resources; it can be successfully implemented even if resources are limited.
► It can be provided in tertiary care facilities, in community health centres and even in children's homes'.

The UK national charity for paediatric palliative care, Together for Short Lives, defines palliative care for children with life-limiting conditions as 'an active and total approach to care, from the point of diagnosis or recognition, embracing physical, emotional, social and spiritual elements through to death and beyond. It focuses on enhancement of quality of life for the child and support for the family and includes the management of distressing symptoms, provision of short breaks and care through death and bereavement'.

---

everyday reality for many. Family carers can experience enormous emotional, physical and financial pressures.[4]

Research suggests that families of children with life-limiting and life-threatening conditions wish for continuous and holistic healthcare, with the option that this is delivered in the home environment.[4] However, most children who die have a pre-existing life-limiting condition,[5] and most die in hospital,[6 7] most frequently in an intensive care environment, where the mode of death is often withdrawal or limitation of life-sustaining treatments.[8-10] The length of stay in the intensive care unit before death is increasing and the costs of hospital care at the end of life are significant.[11 12] This situation presents complex clinical and ethical dilemmas at individual, organisational and societal levels.

Palliative care is an approach to care, which is advocated for all people who live with a life-limiting or life-threatening condition. Current definitions of palliative care are broad (box), which can cause difficulties and lack of clarity for those designing and commissioning specific services.

For clarity, in this paper, children and young people will be referred to throughout as 'children'.

Which children?

There has been a range of previous work concerned with the identification of clinical conditions where palliative care could be beneficial,[13 14] and the following categorisation is provided by Together for Short Lives[15]:

► *Group 1:* life-threatening conditions where access to palliative care services is necessary alongside attempts at curative treatment and/or if treatment fails, such as cancer.

► *Group 2:* conditions such as Duchenne muscular dystrophy, where premature death is inevitable, but where there may be long periods where the child is well.
► *Group 3:* progressive conditions without curative treatment options, such as Batten disease.
► *Group 4:* irreversible but non-progressive conditions, with complex disabilities and healthcare needs which lead to increased likelihood of premature death, such as severe brain injury.

Organisational issues

Currently, there is a wide geographic variation in terms of paediatric palliative care services, and a poorly understood range of commissioning arrangements to support these services. Many services exist as a result of significant contributions from the voluntary sector (including children's hospices), through the efforts of motivated individuals, and through non-recurring funding opportunities rather than the implementation of national policy.[16] A significant development is the emergence of paediatric palliative medicine as a subspecialty of paediatrics.[17 18]

Effective palliative care services for children require strong partnerships between providers, and may require cross-boundary, collaborative commissioning between the statutory and voluntary sectors.[19] In the UK, palliative care for children has specifically been included in national policy, a service specification for paediatric palliative care exists and NICE (National Institute for Health and Care Excellence) Guidelines for end-of-life care for infants, children and young people were published in 2016.[20-24]

Ontology, epistemology and theoretical perspective

Much of the evidence base that guides policy and practice in medicine is derived from experimental research grounded in a positivist paradigm, for example, randomised controlled trials, where a hypothesis can be generated and tested. The positivist approach does not lend itself to research which aims to investigate more complex interventions, such as palliative care, and an interpretive approach is more appropriate. The experience of healthcare services by children with a life-limiting or life-threatening condition and members of their family are shaped and influenced by many interlinked factors including their own personal experiences, values and cultural influences, the values of the healthcare team and the healthcare system, the specific context in which care is delivered and the relationships between those involved in providing and receiving care.[25-27]

The proposed research seeks to understand the mechanisms and influences that shape the experience of care in order to inform both the development and implementation of policy for palliative care services for young children with life-limiting conditions. The methodological approach identified as most appropriate for this research aim is realism, an approach which is increasingly used in healthcare and health sector management research.[28-31]

First described by Bhaskar in the 1970s,[32] and subsequently by Pawson and Tilley [33] realism seeks to understand how phenomena come about as a result of hidden mechanisms, enacted under certain circumstances.[33][34] It acknowledges that there are a wide variety of dynamic contexts and mechanisms which can affect outcomes, including geographical and environmental factors, social and cultural issues and historical factors, and provides a generative approach allowing for the proposal of theories to guide the implementation of policy into practice.[35]

Rationale for research

Despite the range of recommendations for the provision and development of paediatric palliative care services, there remains a lack of research evidence to support the implementation of these guidelines.[1][18][36–38] The proposed research seeks to address this gap using a realist approach to address research questions that correlate with the practical concerns associated with service delivery. The findings and theories that are generated will provide in-depth insights that will be of immediate relevance to clinicians, commissioners and policy makers, as well as to patients and their families.

Research questions

1. How do children with life-limiting and life-threatening conditions and their family members perceive healthcare services, and in particular 'palliative care'?
2. What are the experiences and preferences of children living with a life-limiting or life-threatening condition and/or their families, in relation to the delivery of healthcare services?
3. What are the facilitators and barriers to the delivery of palliative care for children, and how might these be overcome?
4. What should an integrated model of palliative care for children look like?

METHODS AND ANALYSIS

In order to conduct an exploration of the experiences of healthcare from the perspective of children with life-limiting and life-threatening conditions and their families, we will adopt qualitative research methods and a narrative-based approach, suitable for complex, emotionally charged subject areas.[39][40] Active listening, reflection, a flexible approach and insight into the narratives being co-constructed between participant and researcher will be necessary throughout.

This is the protocol for an in-depth longitudinal qualitative study using semistructured interviews with school-aged children (5–18 years) and one or two of their household family members.

Benefits of longitudinal studies include being able to describe the changing needs of the children and their families, and their experience of services, over time,[41] and enabling rapport to build between researcher and participant.

Neonates, preschool children and young people aged over the age of 18 years are excluded from this study. Specific issues around healthcare services arise when considering neonatal care and young people who are making the transition from paediatric to adult services, both of which warrant research in their own right. Research methods would need to be tailored to interview preschool children; this is also an area for potential future research.

The research plan has been informed by review of relevant literature, patient and public involvement (PPI) work and advice from local experts via the West Midlands Paediatric Palliative Care Network.

Sampling and recruitment

Recruitment to a study of this nature depends on many factors, including the clinical condition of the child, conflicting demands on the family's time and the motivation and understanding of their clinical teams. Recruitment began following ethical approval in October 2016 and will continue until January 2018.

The approach to participants is through:
1. direct invitation via their clinical team
2. leaflets and posters displayed in public areas in the hospital (such as notice boards on wards and in outpatients).

The research will be introduced to clinical teams in both the hospital and the community through formal presentations at departmental meetings and to individual clinicians at their request, as well as to the paediatric palliative care network. The researcher, SM, will undertake a period of shadowing with clinical teams, on hospital wards, in outpatient clinics and in the community.

Potential participants will be provided with a participant information sheet, with details of the researcher, the project, how to get involved and a contact telephone number and email address.

Inclusion and exclusion criteria are outlined in table 1. Our aim initially is to purposively sample children so that each of the four Together for Short Lives categories are represented. However, since children live with such individual and highly complex conditions, we anticipate that achieving this may be difficult. The study population will therefore be children with life-limiting or life-threatening conditions, aged from 5 to 18 years, and their family members, some of whom have experience of a palliative care service, and some who do not.

The study has been carefully designed to ensure that all of the children have the opportunity to participate and that wherever possible the views of the child are included, by tailoring each individual interview to their needs and capabilities (including the consent and agreement process). This may include having a learning disability or communication difficulties associated with their condition.

Ethical approval has been granted for the recruitment of 12–14 families to take part in a series of interviews (longitudinal interviews). The aim is to continue

261

Open Access

Table 1	Inclusion and exclusion criteria
Inclusion criteria	1. Children aged 5–18 years (school age) with a life-limiting or life-threatening condition who are under the care of the community children's nursing team and/or the children's hospital and who either: ► receive palliative care services ► are aware of (have had discussions about) palliative care services ► are living with relapsing or refractory disease ► or have had a life-threatening episode (admission to the paediatric intensive care unit).
Exclusion criteria	2. Their family members, who live in the same household.[63] ► Children aged <5 years and >18 years. ► Families of children <5 years and >18 years old. ► Children and families with whom the research team has clinical contact. ► Children and/or families who do not wish to participate. ► Children who are too unwell will not be approached for interview, but their family members may still participate if they wish to. The researcher will take advice from parents about when an individual child is 'too unwell' to take part. ► Children who are unable to participate in a conversational interview for any reason related to their condition will not be approached for interview, but their family members may participate if they wish to. ► Children and families who are unable to provide informed consent in English will not be approached for interview.

to conduct interviews until data saturation is achieved,[42] however, given the uniqueness and individuality of the stories of children and families, it is possible that new themes will continue to emerge such that data saturation is impossible. We will aim for saturation of the main themes that emerge from the data, and identify emergent themes, which may form the basis of future research.

Interview plan

Interviews will be carried out by SM, a researcher who is also a general practitioner (GP) with advanced communication skills training and previous experience in qualitative interviewing. According to the preference of participants, interviews will be conducted with individuals, or with the child and parent together, in their preferred location. One or two family members will be interviewed in each family, either individually or together, depending on their preference and what is most convenient for them.

Interviews will be open and conversational, using a blended approach of interview techniques, with passive interviewing allowing the participant space and time to tell their story (narrative), and more active techniques, including appreciative inquiry, which asks 'What works well?' and 'Why does it work well?',[4 43] employed. A topic guide (table 2) will guide the interview; this will continue to develop iteratively throughout the research, with adaptations made during each interview and in response to each individual participant.

For interviews with children, a range of techniques will be used including depersonalising questions, developing a narrative in the third person, and using props and toys to encourage storytelling. Arts-based activities will be used, where appropriate, as a mutual point of focus for the researcher and participant, or as a focus of the interview, as in the draw–write–tell technique.[44] PPI advice

has been sought on the format of interviews for children (table 3).

Each interview will be audiorecorded, with field notes made to include any additional comments from the child or family made once the audiorecording has stopped,[45] reflections on the interview and observation of the family situation, environment, behaviour and any other interactions that may take place (for example, with other members of the family and clinical staff in hospital or on the phone).

Participants will be asked whether they would like to participate in interviews that will take place over a period of up to 12 months. These are intended to allow the identification of common themes over time and for theories generated through analysis of earlier interviews to be tested out during later interviews.[41] The time intervals between interviews will be individually agreed, depending on the child and family circumstances. The method of communication with each family will also be individually agreed (phone or email). Up to three interviews with each participant are aimed for.

We anticipate practical challenges with conducting longitudinal interviews relating to fluctuations and changes in the clinical condition of each child. Depending on their condition, some children will respond well to treatments and get better. Others may suffer unexpected complications of their condition or treatment, and some may suffer deteriorations which bring about the possibility of dying. To manage the research in this context, we will check the family understanding of the situation before every interview. On occasions, interviews may need to be postponed and rearranged at late notice due to a change in circumstances.

For children who are unable to participate in interviews due to their condition, family members will be

Table 2 Topic guide

For all families	For those aware of 'palliative care'
Introduction	**Palliative care and you (if appropriate)**
Please tell me your story, in any way that you can/want to	► Do you have 'palliative care' services?
► Please tell me the story of you	► Have you ever heard the term 'palliative care'?
► Please can you tell me about you?	► What does that mean to you?
► Your family?	**What do you receive those services for?**
► Your child(ren)	► What do these services provide for you?
What is important to you?	**Does it matter what a service is called?**
► What do you like to do?	**Do you receive services from the hospice?**
► Which places are important to you?	**Can you tell me how you came to receive palliative**
► Where do you spend your time?	**care/know the palliative care nursing team/the**
Which services are involved in your care?	**hospice?**
► Who comes to see you?	► When were you referred?
► What do they do?	► Who brought it up/made the referral?
► What is helpful?	► How was this discussed with you?
► What is not?	► How was that for you/your family?
Which healthcare professionals do you consider to be key in	**Do you think that medical/nursing staff receive enough**
the delivery of your care?	**training in this area?**
► What works best?	► What makes you think that?
► Which services/professionals are most helpful?	**Anything else?**
► Which services/professionals do you value most?	
► What does not work?	
How do you think services could be improved?	
Do you talk to other children/young people/families about your	
healthcare/services?	
► What do you tell your friends?	
► What tends to come up in these discussions?	
► Would you recommend these services to others?	

Questions in bold are leading questions. Bulleted questions are prompts.

interviewed. Children and families are under no obligation to take part in follow-up interviews if they do not wish to. In these cases, and with their consent, data from previous interviews will still be included in the study.

Data analysis

Interviews will be transcribed verbatim, and NVivo used for data handling. Analysis of interview transcripts and field notes will commence alongside data collection, with an initial broad thematic analysis. All data will be coded, and codes grouped into broad overarching themes.

This initial analysis will be followed by an in-depth, narrative analysis, using structure form analysis to examine not just what is being said, but how it is being said, and to propose what works, for whom, in what circumstance at

a micro (immediate clinical team), meso (local organisation) and macro (wider healthcare system) level perspective.[33 46 47]

The collection of longitudinal data allows for innovative approaches to be taken in data analysis.[41] Matrices will be developed to identify key times for families and identification of cross-cutting themes at these times, for example, the time of diagnosis, an admission to intensive care or referral to a palliative care team.

Peer review and respondent validation will take place throughout the data analysis as follows[48 49]:
1. Peer review: SM will code all of the data. A selection of transcripts will be reviewed and independently coded by other members of the research team in order to

Table 3 Feedback from PPI groups on interview plans

January 2016	"Those who are passionate about improving palliative care will take part regardless of how sensitive this may be"
July 2016	"Remember young people who are seriously ill are more mature, they have to grow up"
	"Keep it simple as often a child will openly speak anyway"
	"'Do you talk about it to your friends?' is a good question, a good way to talk to most ages"
	"Children are more eloquent, mature and more capable than you think"
October 2016	"Use pictures and images, more emojis"
February 2017	"Doesn't make me uncomfortable as I think it is very important and relevant"

PPI, patient and public involvement.

263

decrease lone researcher bias.[48] The coding frameworks will be discussed and compared, allowing further development of categories and themes.

2. Respondent validation: by returning to participants to conduct longitudinal interviews, there is an opportunity to check, validate or refute emergent themes from the initial data analysis.

Healthcare professional perspectives

There are 12 paediatric palliative care networks in the UK, which include professionals from a range of organisations within paediatric palliative care. Several have patient and family representatives. Arrangements will be made to present study findings to four of the UK networks at existing meetings. The presentation will be followed by a structured focus group which will aim to first to test out and validate with palliative care professionals the themes from the research findings, and second aims to collect views of professionals. These multiple perspectives will inform and guide the formulation of recommendations for healthcare services in the future.[50]

An expression of interest email will be circulated to network chairs via Together for Short Lives, and arrangements made to attend meetings from networks who respond. Audiorecorded focus group discussions will be carried out at those meetings by SM.

Patient and public involvement

PPI has been integral to the design and conduct of the study. Members of existing groups at a children's hospital and children's hospice have provided advice on the study proposal and design. Smaller groups have been recruited for specific activities, including conference presentations. Group members range in age from 12 to 22 years. PPI activities are outlined in table 4, and will continue throughout the project, with the aim of coproducing the recommendations for the model of care. This will involve structured group sessions during which anonymised findings of the data analysis will be presented to the group for feedback and comment. A patient experience framework will be used to structure the discussion and to build recommendations.[51]

Strengths and limitations

The strength of this study lies in the in-depth, contextual qualitative nature of the data, with multiple child and family member stories captured over time. Our anticipated study population is diverse in terms of age, clinical condition, cultural background and family structure, allowing detailed consideration of the role of healthcare services in effectively recognising and supporting children and families with their individual needs. All of the children and families included in the study could benefit from palliative care as it is currently defined,[15] however, not all will have had conversations about this with their clinicians, or been referred to specialist palliative care services. Given the nature of their clinical conditions, including for some the inability to communicate verbally or deterioration in their health, recruitment and retention within the study is likely to become a challenge and will require a reflexive, flexible approach.

Potential limitations in the study include our exclusion of neonates, preschool children and young people at transition (over the age of 18 years). These groups all warrant research in their own right. Given the time and resource constraints of the study, all interviews will be carried out in English. Further research into the experiences of children and families who cannot communicate in English is necessary. There will be ongoing consideration of sources of bias. Recruitment bias is being addressed by aiming for a diverse sample and providing access to project information independent of the clinical teams. Data saturation will be sought during data analysis, with an ongoing process of reflection and peer review to address any possible unconscious bias of the researcher (SM).

Table 4	PPI activities
Completed PPI activities	► Developing the original research proposal. ► Advising on the language used in the study (suggesting a change in the title from 'Palliative Care for Children and Young People: What? When? How?' to 'The Journey through Care'. ► Developing participant resources including leaflets for older and younger children. ► Interview design, including suggesting how questions could be phrased and asked. ► Providing family perspectives to a literature review, and becoming a coauthor on the paper.[18] ► Taking part in oral presentations at regional conferences.
Work in progress	► Designing conference posters and presentations for national conferences. ► Working as coresearchers to carry out a survey study to investigate understanding of the term 'palliative care' for children and young people and healthcare professionals.
Future plans	► Working with a project-specific group to explore the findings of the research study and develop recommendations for a new model of care. ► Dissemination projects including conference presentations, posters, website design, use of social media, infographics and films.

PPI, patient and public involvement.

264

Figure 1 Ethical issues in longitudinal qualitative research for children and families in palliative care.

Ethics and dissemination

Research with children raises ethical and legal considerations around recruitment, consent and data collection.[52][53] In addition, research regarding palliative and end-of-life care can be emotionally demanding and distressing for those involved. There are also particular ethical issues to consider given the longitudinal nature of the study.[54]

We are recruiting children and families who are potentially vulnerable and may be experiencing considerable distress. The justification for our approach is that children and their families in this situation are rarely asked about their experiences, but talking to them and understanding their experiences is essential in order to be able to design and develop services that respond to their actual needs. Here, we summarise our approach to the ethical issues the study raises (figure 1).

Language

Published literature suggests that the term 'palliative care' is poorly understood and perceived negatively,[55–59] a view confirmed by our PPI group. The scope of our study is therefore to investigate the experiences of children with 'life-limiting', 'life-threatening' and 'conditions which may or may not get better', whether or not they have heard of palliative care or receive care from specialist services. 'Palliative care' will be avoided in participant information sheets and interviews, unless individuals are already familiar with palliative care services or bring it up themselves.

Recruitment

There is an ethical challenge in terms of potential coercion to the study by clinicians who know the family well. Clinicians will therefore only provide study information but will not actively recruit families; the initial expression of interest is from the family to the researcher. The researcher (SM) will then discuss the study in person or by phone with the child and their family member(s) and answer any questions before arranging a time for interview. Participants will be made aware that they can decline to take part or to withdraw at any stage without having to give a reason. Interviews are only carried out at a time that is mutually agreed and minimises any potential inconvenience or intrusion.

Equity of access to the study

Recruitment through clinical teams is widely used in palliative care research but may be limited by 'gatekeeping'.[60] There may also be families who wish to participate who do not find out about the study through their clinical team. In order to address this, we have designed posters for display on hospital wards and in outpatients, and at the local children's hospice, and a paragraph for organisational newsletters. These provide the direct contact details for the researcher (email, text or phone).

Open Access

Table 5 Planned outputs from the research

Academic/clinical audiences	Patient, public and policy maker audiences
► PhD thesis	► A report prepared for participants and PPI volunteers.
► Peer-reviewed publications	► The development of guidance for commissioners and providers.
► Presentations at national and international conferences	► The development of resources that are accessible to patients and families.

The study setting is Birmingham, UK, a city where the population is highly diverse in terms of family situation and multiculturalism. Over 50% of families with a child known to palliative care services in Birmingham and Solihull are from black or minority ethnic backgrounds.[61] Many of these families speak English as a first or second language, so within the time and resource constraints of this study, interviews will be carried out with those who can provide informed consent and take part in an interview in English.

Consent

Consent for the study raises ethical and legal issues with children who are under the age of 16 years and/or do not have the capacity to consent. We will aim for written and/or verbal consent and agreement from every individual for every interview.

For children under the age of 16, written consent will be obtained from the parent and then verbal or written agreement obtained from the child.

In keeping with the Mental Capacity Act, there is an assumption of capacity in young people aged 16 years and over, so they will be asked for consent first, followed by agreement from their parent(s). Parental agreement is not a legal requirement, but conducting an interview with a young person about a potentially difficult subject without the knowledge or agreement of their parents is an ethical concern. If there is a concern that the young person lacks capacity or is considered particularly vulnerable, for example, with a learning disability, parents will be asked to provide verbal and written consent in addition to the young person's agreement.

Parental consent is required for any interview to be carried out in the family home.[53]

For a child on a full care order, social worker consent would replace that of parental consent. Where possible parental consent/agreement will also be sought.

Interviews

Subject areas discussed during interviews may cause distress to participants, and recruitment may occur soon after sensitive conversations. We have designed the study to ensure that the risks and burden associated with taking part in the study are minimal.

Qualitative interviews will be informal and reflexive to the needs of the participant. In the event that a participant experiences any difficulties during the interview, such as tiredness or distress, the interview will be halted, and if necessary brought to an end. Adequate time will be given for debrief, and the researcher will provide information about local resources for support if necessary. Interviews will be carried out at a time and in a location that is convenient to the participants. If this is in hospital, the researcher (SM) will liaise closely with clinical teams so that the research does not interfere with routine clinical care and ward work.

Longitudinal interviews

Family views and understanding of what might happen next as a result of the condition of the child will be discussed sensitively, and any follow-up interviews scheduled around possible further treatments. If it seems likely that there will be a deterioration in the condition of the child, this is explored carefully and an agreement made with the individual family about whether they want to continue to participate in the study.

Anonymity and confidentiality

All interview data will be anonymised with personal identifiers removed. Any qualitative interview data that could identify child, families or any professionals involved in their care because of the individuality and context of the narrative is included in the data analysis, but will be excluded from reporting.

Field notes and anonymised interview transcripts will be stored securely on a password protected university hard drive.

Minimising harm to the researcher

There is a need for clearly defined boundaries for a researcher–participant relationship in a longitudinal study of this type. It will be made clear to participants at the time of consent that it is not the role of the researcher to provide personal support or clinical advice. With the risk of emotional distress for the researcher, plans to ensure adequate support through regular academic supervision and access to a counsellor are in place.

Serious concerns and safeguarding

If information contained in a participant's response indicates a serious clinical or safeguarding concern or an issue which may jeopardise the safety of the participant or another person, this will be escalated appropriately in line with the protocols of the community or hospital trusts. This may on very rare occasions necessitate a breach of participant confidentiality in order to maintain their safety. Participants will be informed of any disclosure and to whom it is made.

266

Dissemination plan

The research is embedded in plans for impact. Table 5 outlines our planned outputs. We will work on traditional academic and clinical outputs, including manuscripts with the results of the study for publication in a peer-reviewed journal. Simultaneously work will be carried out with the PPI group to plan innovative, accessible outputs for patients, the public and commissioners which will include infographics and film based reports outlining our recommendations.

REFERENCES

1. Fraser LK, Miller M, Hain R, et al. Rising national prevalence of life-limiting conditions in children in England. *Pediatrics* 2012;129:e923–29.
2. Fraser LK, Parslow RC, McKinney PA, et al. Life-limiting and life-threatening conditions in children and young people in the United Kingdom. 2012;129:1–7.
3. Office for National Statistics. *Mortality statistics: deaths registered*, 2007.
4. Coad J, Kaur J, Ashley N, et al. Exploring the perceived met and unmet need of life-limited children, young people and families. *J Pediatr Nurs* 2015;30:45–53.
5. Sidebotham P, Fraser J, Fleming P, et al. Patterns of child death in England and Wales. *Lancet* 2014;384:904–14.
6. Confidential Enquiry into Maternal and Child Health. *Why children die: a pilot study*. London: CEMACH, 2008.
7. Department of Health (2007). *Palliative care statistics for children and young adults*. London: Department of Health, 2007.
8. McCallum DE, Byrne P, Bruera E. How children die in hospital. *J Pain Symptom Manage* 2000;20:417–23.
9. Ramnarayan P, Craig F, Petros A, et al. Characteristics of deaths occurring in hospitalised children: changing trends. *J Med Ethics* 2007;33:255–60.
10. Epstein D, Brill JE. A history of pediatric critical care medicine. *Pediatr Res* 2005;58:987–96.
11. Widger K, Seow H, Rapoport A, et al. Children's end-of-life health care use and cost. *Pediatrics* 2017;139:e20162956.
12. Plunkett A, Parslow RC. Is it taking longer to die in paediatric intensive care in England and Wales? *Arch Dis Child* 2016;101:798–802.
13. Hain R, Devins M, Hastings R, et al. Paediatric palliative care: development and pilot study of a 'Directory' of life-limiting conditions. *BMC Palliat Care* 2013;12:43.
14. Noyes J, Edwards RT, Hastings RP, et al. Evidence-based planning and costing palliative care services for children: novel multi-method epidemiological and economic exemplar. *BMC Palliat Care* 2013;12:18.
15. Together for short lives. Children's palliative care definitions. http://www.togetherforshortlives.org.uk/assets/0000/4089/CPC_definitions.pdf
16. MacNamara-Goodger K, Feudtner C. History and epidemiology. In: Goldman A, Hain R, Liben S, eds. *Oxford textbook of palliative care for children*. 2nd edn. Oxford: Oxford University Press, 2012.
17. Hain R, Heckford E, McCulloch R. Paediatric palliative medicine in the UK: past, present, future. *Arch Dis Child* 2012;97:381–4.
18. Mitchell S, Morris A, Bennett K, et al. Specialist paediatric palliative care services: what are the benefits? *Arch Dis Child* 2017;102:923–9.
19. Rowse V. Home-based palliative care for children: the case for funding. *Paediatr Nurs* 2006;18:20–4.
20. National Institute for Health and Care Excellence (NICE). End of life care for infants, children and young people with life-limiting conditions: planning and management. https://www.nice.org.uk/guidance/ng61
21. Villanueva G, Murphy MS, Vickers D, et al. End of life care for infants, children and young people with life limiting conditions: summary of NICE guidance. *BMJ* 2016;355:i6385.
22. Scottish Government. Strategic framework for action on palliative and end of life care. 2015 http://www.gov.scot/Topics/Health/Quality-Improvement-Performance/peolc/SFA
23. Welsh Government. End of life care delivery plan. 2014 http://gov.wales/topics/health/nhswales/plans/end-of-life-care/?lang=en
24. Department of Health, Social Services and Public Safety, Northern Ireland. Living matters, dying matters: a palliative and end of life care strategy for adults in Northern Ireland. www.dhsspsni.gov.uk/publications/living-matters-dying-matters-strategy-2010
25. Mason J. *Qualitative Researching*. London: SAGE Publishing, 2002.
26. Crotty M. *The foundations of social research: meaning and perspective in the research process*. Sydney: Allen & Unwin, 1998.
27. Ormston R, Spencer L, Barnard M.The foundations of qualitative research. In: Ritchie J, Lewis J, McNaughton Nicholls C, Ormston R, et al. eds. *Qualitative research practice*, 2013.
28. Schiller CJ. Critical realism in nursing: an emerging approach. *Nurs Philos* 2016;17:88–102.
29. Walsh D, Evans K. Critical realism: an important theoretical perspective for midwifery research. *Midwifery* 2014;30:e1–e6.
30. Connelly J. A realistic theory of health sector management. The case for critical realism. *J Manag Med* 2000;14:262–71.
31. Ragin C, Becker H. *What is a case?Exploring the foundations of social inquiry*. Cambridge University Press, 1992.
32. Bhaskar R. *A realist theory of science*. Brighton: Harvester, 1975.
33. Pawson R, Tilley N. *Realistic evaluation*. London: Sage Publications, 1997.
34. Clark AM, MacIntyre PD, Cruickshank J. A critical realist approach to understanding and evaluating heart health programmes. *Health* 2007;11:513–39.
35. Sayer A. *Realism and social science*. London: Sage Publications, 2000.
36. Hain RD. Palliative care in children in Wales: a study of provision and need. *Palliat Med* 2005;19:137–42.
37. Hain RW. Palliative care in children: Who needs it, and why? Acta *Paediatrica*. , 2011:100, 4.
38. Baker JN, Levine DR, Hinds PS, et al. Research priorities in pediatric palliative care. *J Pediatr* 2015;167:467–70.
39. Akard TF, Gilmer MJ, Friedman DL, et al. From qualitative work to intervention development in pediatric oncology palliative care research. *J Pediatr Oncol Nurs* 2013;30:153–60.
40. Kendall M, Harris F, Boyd K, et al. Key challenges and ways forward in researching the "good death": qualitative in-depth interview and focus group study. *BMJ* 2007;334:521.
41. Murray SA, Kendall M, Carduff E, et al. Use of serial qualitative interviews to understand patients' evolving experiences and needs. *BMJ* 2009;339:b3702.
42. Gill P, Stewart K, Treasure E, et al. Methods of data collection in qualitative research: interviews and focus groups. *Br Dent J* 2008;204:291–5.
43. Carter B, Coad J, Goodenough T. Department of Health. *Community Children's Nursing in England: An appreciativereview of CCNs*, 2009.

267

http://www.dh.gov.uk/en/Publicationsandstatistics/Publications/Publi cationsPolicyAndGuidance/DH_1248-98

44. Greig A, Taylor J, MacKay T. *Doing research with children. A practical guide*. London: SAGE, 2008:113–20.

45. Coad J, Gibson F, Horstman M, *et al*. Be my guest! Challenges and practical solutions of undertaking interviews with children in the home setting. *J Child Health Care* 2015;19:432–43.

46. Lieblich A, Tuval-Mashiach R, Zilber T. *Narrative research: reading, analysis and interpretation*. London: Sage, 1998.

47. Bingley AF, Thomas C, Brown J, *et al*. Developing narrative research in supportive and palliative care: the focus on illness narratives. *Palliat Med* 2008;22:653–8.

48. Burnard P, Gill P, Stewart K, *et al*. Analysing and presenting qualitative data. *Br Dent J* 2008;204:429–32.

49. Burnard P. A method of analysing interview transcripts in qualitative research. *Nurse Educ Today* 1991;11:461–6.

50. Kendall M, Murray SA, Carduff E, *et al*. Use of multiperspective qualitative interviews to understand patients' and carers' beliefs, experiences, and needs. *BMJ* 2009;339:b4122.

51. Staniszewska S, Boardman F, Gunn L, *et al*. The warwick patient experiences framework: patient-based evidence in clinical guidelines. *Int J Qual Health Care* 2014;26:151–7.

52. Medical Research Council. *MRC Ethics Guide: Medical research involving children*. London: Medical Research Council, 2004. www.mrc.ac.uk

53. Shaw C, Brady L-M, Davey C. *Guidelines for research with children and young people*. London: National Children's Bureau, 2011.

54. Calman L, Brunton L, Molassiotis A. Developing longitudinal qualitative designs: lessons learned and recommendations for health services research. *BMC Med Res Methodol* 2013;13:1–10.

55. Hui D, De La Cruz M, Mori M, *et al*. Concepts and definitions for 'supportive care' 'best supportive care' 'palliative care' and 'hospice care' in the published literature, dictionaries, and textbooks. *Support Care Cancer* 2013;21:659–85.

56. Pastrana T, Jünger S, Ostgathe C, *et al*. A matter of definition–key elements identified in a discourse analysis of definitions of palliative care. *Palliat Med* 2008;22:222–32.

57. Twamley K, Craig F, Kelly P, *et al*. Underlying barriers to referral to paediatric palliative care services: knowledge and attitudes of health care professionals in a paediatric tertiary care centre in the United Kingdom. *J Child Health Care* 2014;18:19–30.

58. Monterosso L, Kristjanson LJ. Supportive and palliative care needs of families of children who die from cancer: an Australian study. *Palliat Med* 2008;22:59–69.

59. Morstad Boldt A, Yusuf F, Himelstein BP. Perceptions of the term palliative care. *J Palliat Med* 2006;9:1128–36.

60. Crocker JC, Beecham E, Kelly P, *et al*. Inviting parents to take part in paediatric palliative care research: a mixed-methods examination of selection bias. *Palliat Med* 2015;29:231–40.

61. Hunt A, Coad J, West E, *et al*. *The big study for life-limited children and their families: final research report*. Bristol: Together for Short Lives, 2013.

62. World Health Organisation. Cancer: WHO definition of palliative care. http://www.who.int/cancer/palliative/definition/en/

63. Office for National Statistics. *2011 census glossary of terms office for National Statistics*, 2014.

268

BMJ Open

The journey through care: study protocol for a longitudinal qualitative interview study to investigate the healthcare experiences and preferences of children and young people with life-limiting and life-threatening conditions and their families in the West Midlands, UK

Sarah Mitchell, Anne-Marie Slowther, Jane Coad and Jeremy Dale

Updated information and services can be found at:
http://bmjopen.bmj.com/content/8/1/e018266

These include:

References	This article cites 39 articles, 10 of which you can access for free at: **http://bmjopen.bmj.com/content/8/1/e018266#ref-list-1**
Open Access	This is an Open Access article distributed in accordance with the terms of the Creative Commons Attribution (CC BY 4.0) license, which permits others to distribute, remix, adapt and build upon this work, for commercial use, provided the original work is properly cited. See: http://creativecommons.org/licenses/by/4.0/
Email alerting service	Receive free email alerts when new articles cite this article. Sign up in the box at the top right corner of the online article.
Topic Collections	Articles on similar topics can be found in the following collections Palliative care (82)

Notes

QUALITY IMPROVEMENT

Ethics and patient and public involvement with children and young people

Sarah Jane Mitchell,[1] Anne-Marie Slowther,[1] Jane Coad,[2] Shazaan Akhtar,[3] Elizabeth Hyde,[4] Dena Khan,[4] Jeremy Dale[1]

[1]Warwick Medical School, University of Warwick, Coventry, UK
[2]Faculty of Health and Life Sciences, Coventry University, Coventry, UK
[3]Birmingham Children's Hospital Young Persons Advisory Group, Birmingham Children's Hospital, Birmingham, UK
[4]NIHR CRN Young People's Steering Group, Stafford, UK

Correspondence to
Dr Sarah Jane Mitchell, Warwick Medical School, University of Warwick, Coventry CV4 7AL, UK;

ABSTRACT

Patient and public involvement (PPI) is important both in research and in quality improvement activities related to healthcare services . While PPI activities do not require formal ethical approval, they can raise a number of ethical concerns, through the introduction of complex technical medical concepts, challenging language or sensitive subject areas. There is very little published literature to guide ethical practice in this area. We have been conducting PPI with children and young people throughout a research study in paediatric palliative care. PPI started during the application process and continued to guide and shape the research as it progressed. Ethical issues can arise at any time in PPI work. Although many can be predicted and planned for, the nature of PPI means that researchers can be presented with ideas and concepts they had not previously considered, requiring reflexivity and a reactive approach. This paper describes how we considered and addressed the potential ethical issues of PPI within our research. The approach that emerged provides a framework that can be adapted to a range of contexts and will be of immediate relevance to researchers and clinicians who are conducting PPI to inform their work.

BACKGROUND

Patient and public involvement (PPI) is the active involvement of patients and members of the public in the design and process of research. It aims to ensure that research is relevant to the intended audience and that their views are taken into account.[1] The importance of the active participation of children and young people in research that concerns their care is increasingly recognised[2 4] and is a specific focus of INVOLVE, the UK national advisory group for the advancement and promotion of public involvement.[5 6] PPI can take place at any stage of the research process, from the development of the initial research questions through to specific aspects of study design, including data analysis and dissemination. It is required for many research grant applications. PPI is also an important element of service design projects in healthcare.[7 8]

Researchers, clinicians and healthcare managers have a responsibility to ensure that PPI is not tokenistic.[9 10] Those who conduct PPI for research, service design or quality improvement must be willing to listen to and act on ideas and suggestions from children and young people, including any that challenge their own ideas or those that they have not considered before.

In the UK, there is no requirement for ethical approval when undertaking PPI work. Guidance for the conduct of PPI includes the Royal College of Paediatrics and Child Health (RCPCH) research charter and resources from INVOLVE.[11–13] There are also PPI reporting frameworks such as the Guidance for Reporting Involvement of Patients and the Public (GRIPP) checklist[14] and the Public Involvement Impact Assessment Framework (PiiAF),[15] a tool to assess the impact of PPI. A range of guidance on ethical research with children is available,[5 16 17] but little specifically relates to the ethics of PPI with children and young people and the need for an ethical code of conduct for PPI has been raised.[18]

OBJECTIVE

The purpose of this paper is to describe an ethical framework to guide PPI that was developed for paediatric palliative care research.

All photographs are included with the consent of the young people who are pictured.

AN INTRODUCTION TO THE PPI GROUPS

Our PPI has been with existing groups of children and young people at a children's

Check for updates

hospital (Birmingham Children's Hospital Young Person's Advisory Group and the National Institute for Health Research Clinical Research Network Young Person's Advisory Group) and a children's hospice (Acorns Children's Hospice, West Midlands). Group members range in age from 12 to 20 years. Accessing existing groups from a range of organisations had several benefits; members have a wide range of experience and have already received training in elements of research, policy and communication. Some have previous experience in both research and service design projects. The groups are supported by research nurses and support workers, and our PPI work has been incorporated into an established timetable for group meetings that is convenient for young people.

From these groups, we have recruited smaller groups for specific activities, including conference presentations, writing papers[19] and the development of further research.

ETHICAL PPI IN PAEDIATRIC PALLIATIVE CARE RESEARCH: A PRACTICAL EXAMPLE AND A FRAMEWORK

Research in paediatric palliative care is a sensitive subject area. This paper describes our approach to PPI with children and young people for research in paediatric palliative care , during which we constructed a framework for an ethical approach to guide our PPI. The framework has wider relevance to other research or quality improvement projects (table 1). An ethically sound approach will add quality to all PPI activity, whether that is in research, service design or quality improvement.

Prioritise PPI with children and young people

Article 12 of the United Nations Convention on the Rights of the Child states that all children and young people who are capable of forming their own views have a right to express those views freely in all matters affecting them, with the views of the child being given due weight in accordance with their age and maturity.[5] However, there is wide debate among academics regarding the autonomy of children and young people. Some argue that children and young people are competent and able to make their own decisions. Others are more cautious, arguing that competence is difficult to assess in children as their level of maturity and experience varies significantly depending on the context.[20]

Either way, children and young people want their contribution to have impact.[1 21 22] Before starting PPI, adult researchers must be clear about how they will accommodate suggestions that are made. This can be challenging,[4] particularly as the perspectives of children may differ from their own. There are inevitable power dynamics between the adult researcher and PPI group members during each interaction. The adult researcher comes with qualifications and professional status.[23] Children and young people bring their own knowledge and experience of the world and can challenge clinicians and reseachers who are used to assuming responsibility for the management and coordination of teams in their work.[24]

In order to build an appropriate rapport with the group and to fulfil the objectives of the PPI, the researcher requires insight into these power dynamics, good communication skills, self-awareness and a reflexive approach in order to move between different roles: teacher, colleague, mentor and group facilitator, depending on the task. Understanding the experience and knowledge, ambition, qualifications and status of PPI group members in relation to each other also helps and can develop naturally over a number of sessions with the same group.

In our PPI, children and young people have presented views and experience whcih have challenged the researcher including their knowledge of technology and social media. Some bring the experience of illness as a sibling or as a patient. They have strong views about aspects of the research project including the language used and the design of study resources and interviews, and they are passionate about dissemination and impact. The established groups we have been working with already have an identity and autonomy as a group, and high expectations of how their views are valued. This may have been different if the group had been made up of volunteers selected more generally.

In order to assure group members that their contributions have resulted in changes, we provide regular feedback at subsequent meetings, through sharing documents such as participant information resources and through a project Twitter account (@journeythrucare), set up at the group's request.

Agree language and work towards shared understanding of tasks

A crucial aspect of communication in PPI is ensuring shared understanding for each particular task. Anticipating this challenge and planning PPI group sessions with clear, focused questions and tasks are beneficial, and there are suggestions to guide this process in the

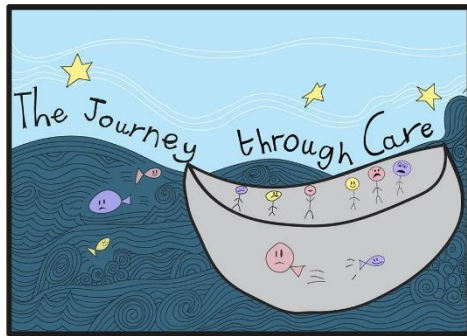| Table 1	An ethical approach to patient and public involvement (PPI): key principles
Step	**Ethical approach**
1	Prioritise PPI with children and young people
2	Agree language and work towards a shared understanding of tasks
3	Gain consent for PPI
4	Maximise the benefits for PPI group members
5	Minimise the risk of harm
6	Ensure equity of access to PPI
7	Provide training for the researcher
8	Offer training for the PPI group
9	Provide funding and recognition

Figure 1 Project logo.

RCPCH&Us Recipes for Engagement resource.[25] Clear explanations of how each task relates to the research and regularly checking back with group members to ensure there is shared understanding of every PPI task are helpful strategies.

Group members question medical jargon and terms that are unfamiliar to them. An example from our work relates to the term 'palliative care'. During our initial PPI activity, group members including young people who had been patients at the local children's hospice, raised concerns about the term 'palliative care'. They had not heard of it, and they perceived it negatively. They associated the term with death and dying and did not want to relate it to themselves or to other children and young people. Similar concerns have been identified in the previous research.[26-28] The young people suggested changing the study title to 'The Journey through Care', designed a logo for the study (figure 1), and participant information leaflets were edited to refer to 'life-limiting' and 'life-threatening' conditions or 'conditions which may or may not get better', but not specifically to 'palliative care'. This has had positive implications for the research and has led us to design further research into language in palliative care.

Gain informed consent

Ethical approval and the use of consent or agreement forms for children, young people or their parents are not necessary for PPI. However, the principles of informed consent should still be carefully considered particularly if the subject of the research is potentially sensitive, when PPI groups include members who may have mixed personal experiences and where group members are under the age of 16 years.

At each stage of our PPI work, we have asked participants for verbal agreement in relation to each activity. Our approach and learning points are as follows:
1. To find out about the size of the group and age range and whether anyone has personal experiences that may be difficult for them before the meeting.

2. To carefully prepare each session in advance, with specific information, tasks and questions for the group in clear, accessible plain English.
3. To allow time for introductions and discussions about any experiences related to palliative care early on in the meeting.
4. Making clear to group members that they are under no obligation to take part in any element of the PPI work for this project and can leave the session at any time.
5. Asking for verbal agreement from group members at each meeting, reminding them about the subject area and confirming that they are all feeling okay to talk about the research.
6. Providing written information to take away, including details of the study and the contact details of the researcher.
7. Encouraging group members, particularly members under the age of 16 years, to discuss their involvement with their parents.
8. Reassuring group members that their contribution can remain anonymous if that is their wish.

Maximise the benefits for PPI group members

The benefits of PPI for those who take part are often listed as contributing to an original piece of research, gaining new skills and knowledge, working with others and having the opportunity to take an active role in dissemination. We have actively sought to provide opportunities for PPI group members to engage in activities that they consider to be beneficial to themselves, as well as of benefit to the study. This has included conference presentations and co-authorship of papers (figure 2).[19]

Minimise the risk of harm

There is a potential for harm to people who take part in PPI related to the discussion of sensitive subject areas. Anticipating and planning this is helpful for both the researcher and group members, providing the security of a clear framework with which to approach problems if they arise.

Figure 2 Patient and public involvement group members at the Royal College of Paediatrics and Child Health conference 2017.

3

Table 2 Patient and public involvement (PPI) risk assessment

Stage of PPI	Potential risks	Plan to mitigate risks
Approaching groups of young people	Young people who live with life-limiting or life-threatening conditions are at constant risk of a deterioration in their health. These are situations that other young people may not be familiar with and which they therefore might find difficult.	Groups of young people, some of whom have life-limiting conditions and others who do not, were approached separately and through two different organisations (the hospital and hospice). PPI work with each group is carried out concurrently but not at the same group sessions.
Support for PPI group members	Discussion of experiences of life-limiting conditions can cause distress for children and young people.	Should any children or young people require support either during or following the meeting, this is provided by the researcher or by the group facilitators. While PPI work can result in open and honest conversations about difficult topics, this is not the same as professional emotional support. If necessary, group members can be provided with information about whether to access further support.
Using research findings to develop recommendations	There are ethical considerations for the research study participants in terms of ensuring their anonymity and to avoid misinterpretation of qualitative data.	The researcher will be responsible for conducting the data analysis before this is shared with PPI group members and for ensuring that findings remain in context. No identifiable or raw data will be shared.

Given the potential risk of harm to PPI group members through conversations about palliative care, our approach to PPI included a brief risk assessment as described in table 2:

We used a method based on 'Tell Me…' from the RCPCH&Us Recipes for Engagement[25] to gather confidential feedback from PPI group members about their personal experiences of being involved in this research. Although we had planned for potential risks and possible distress, this has not been a problem in our study and feedback to date has been very positive.

Ensure equity of access to PPI

There is more that could be done to ensure equitable access to involvement opportunities for all children and young people. This is the focus of national campaigns In the UK including 'OK to ask'[29] and 'I Am Research'.[30] Social media is a powerful tool through which opportunities could be advertised or through which PPI could be conducted.

Through working with groups from both the hospital and the hospice, we have had PPI input from children and young people with a diverse range of family and illness experiences and cultural backgrounds. The groups are advertised widely and children and young people apply to join. Within the PPI, we are also aware of the need for equity of access to opportunities such as attendance at conferences and co-authoring papers. The young people involved in our study have suggested approaching schools and youth groups to present work to a wider audience and to improve awareness and understanding of both research and PPI, through educational sessions and assemblies.

Design training for the researcher

Currently, there is no consistent or standardised training in the conduct of PPI for researchers.[31] Individual researchers need to address their own training needs. This includes the practical skills necessary for effective PPI (such as facilitation of workshops with CYP of different ages) and consideration of the more subtle, complex issues that can arise in PPI including planning for potential ethical concerns. There is work in progress to develop standards for PPI; training resources would support their implementation.[32]

Design training for the group

Children and young people may require access to training that is relevant to the proposed PPI activities. The approach will depend on the activity that PPI group members are being asked to carry out.[33] Researchers are often in a position to provide such training. For example, Coad and her team provided specific training to young people so that they were in a position to take an active role in qualitative data analysis.[33] Other researchers have recruited and trained PPI co-researchers but describe significant challenges regarding governance and administration processes.[34]

PPI can create the opportunity for children and young people to design and carry out their own research which should also be supported with adequate training. Young person to young person research may enable valuable insights that might not emerge in adult to young person research.[35 36]

Provide funding and recognition

PPI work involves time and other costs which should be covered by the cost of the research. Detailed guidance exists for this.[37] The contribution of PPI group members can also be recognised individually and collectively, through thank you certificates, vouchers and events that can be designed in partnership with the PPI group.

4

CONCLUSION

The importance of incorporating PPI with children and young people in research, service design and quality improvement activities is well recognised. However, there has been a little guidance about how best to conduct such activity. This paper describesa systematic approach to recognising and addressing the ethical issues relevant to the planned PPI work for a palliative care researcg project. The framework that emerged is applicable in numerous contexts and c could be used to identify and address ethical concerns pre-emptively, minimisinge the risk of harm to children and young people while maximising the value of their contribution.

REFERENCES

1 Brett J, Staniszewska S, Mockford C, et al. A systematic review of the impact of patient and public involvement on service users, researchers and communities. *Patient* 2014;7:387–95.

2 Coad JE, Shaw KL. Is children's choice in health care rhetoric or reality? A scoping review. *J Adv Nurs* 2008;64:318–27.

3 UNICEF. The United Nations convention on the rights of the child. 1990 http://www.unicef.org.uk/Documents/Publication-pdfs/UNCRC_PRESS200910web.pdf

4 Bird D, Culley L, Lakhanpaul M. Why collaborate with children in health research: an analysis of the risks and benefits of collaboration with children. *Arch Dis Child Educ Pract Ed* 2013;98:42–8.

5 Shaw C, Brady L-M, Davey C. *Guidelines for research with children and young people*. London: National Children's Bureau, 2011.

6 UNICEF.http://www.invo.org.uk/about-involve/:http://www.invo.org.uk

7 Robertson S, Pryde K, Evans K. Patient involvement in quality improvement: is it time we let children, young people and families take the lead? *Arch Dis Child Educ Pract Ed* 2014;99:23–7.

8 Bedford Russell AR, Passant M, Kitt H, et al. Engaging children and parents in service design and delivery. *Arch Dis Child* 2014;99:1158–62.

9 Weil LG, Lemer C, Webb E, et al. The voices of children and young people in health: where are we now? *Arch Dis Child* 2015;100:915–7.

10 Rahimzadeh V, Bartlett G, Longo C, et al. Promoting an ethic of engagement in pediatric palliative care research. *BMC Palliat Care* 2015;14:50.

11 Research and Us: infants', children's and young people's child health research charter. 2016 http://www.rcpch.ac.uk/improving-child-health/research-and-surveillance/infants-children-and-young-people%E2%80%99s-research-charte

12 Bate J, Ranasinghe N, Ling R, et al. Public and patient involvement in paediatric research. *Arch Dis Child Educ Pract Ed* 2016;101:158–61.

13 INVOLVE. Involving children and young people. http://www.invo.org.uk/find-out-more/how-to-involve-people/involving-children-and-young-people/

14 Staniszewska S, Brett J, Mockford C, et al. The GRIPP checklist: strengthening the quality of patient and public involvement reporting in research. *Int J Technol Assess Health Care* 2011;27:391–9.

15 Popay J, Collins M. *The Public Involvement Impact Assessment Framework (PiiAF): executive summary*. Lancaster: University of Lancaster, 2014.

16 Graham A, Powell M, Taylor N. Ethical research involving children. Putting the evidence into practice. *Family Matters* 2015;96:23–8.

17 Morrow V, Richards M. The ethics of social research with children: an overview. *Child Soc* 1996;10:90–105.

18 Pandya R, Bates A. Developing a code of ethics for patient and public involvement in research design. INVOLVE Conference. 2010 http://www.invo.org.uk/posttypeconference/developing-a-code-of-ethics-for-patient-and-public-involvement-ppi-in-research-design/

19 Mitchell S, Morris A, Bennett K, et al. Specialist paediatric palliative care services: what are the benefits? *Arch Dis Child* 2017;102:923–9.

20 Greene S, Hill M. Researching children's experience: methods and methodological issues. In: Greene S, Hogan D, eds. *Researching children's experience*. London: SAGE, 2005:8–9.

21 Brett J, Staniszewska S, Mockford C, et al. Mapping the impact of patient and public involvement on health and social care research: a systematic review. *Health Expect* 2014;17:637–50.

22 Mockford C, Staniszewska S, Griffiths F, et al. The impact of patient and public involvement on UK NHS health care: a systematic review. *Int J Qual Health Care* 2012;24:28–38.

23 Cosby KS, Croskerry P. Profiles in patient safety: authority gradients in medical error. *Acad Emerg Med* 2004;11:1341–5.

24 Nugus P, Greenfield D, Travaglia J, et al. How and where clinicians exercise power: interprofessional relations in health care. *Soc Sci Med* 2010;71:898–909.

25 Royal College of Paediatrics and Child Health. Recipes for engagement. http://www.rcpch.ac.uk/sites/default/files/user31705/%26Us%20Recipes%20for%20Engagement_IF_A5_AW_ONLINE.pdf2016

26 Hui D, De La Cruz M, Mori M, et al. Concepts and definitions for "supportive care," "best supportive care," "palliative care," and "hospice care" in the published literature, dictionaries, and textbooks. *Support Care Cancer* 2013;21:659–85.

27 Pastrana T, Jünger S, Ostgathe C, et al. A matter of definition– key elements identified in a discourse analysis of definitions of palliative care. *Palliat Med* 2008;22:222–32.

28 Morstad Boldt A, Yusuf F, Himelstein BP. Perceptions of the term palliative care. *J Palliat Med* 2006;9:1128–36.

274

29 NIHR. OK to ask. 2015 http://www.research.hscni.net/ok-ask-campaign

30 NIHR. IAm research. 2017 http://www.nihr.ac.uk/news-and-events/support-our-campaigns/i-am-research/2017

31 Laurie L, Wellings A. A. training and support for ppi in research: what have your experiences been? INVOLVE conference. 2012.

32 Public involvement standards development. 2017 https://sites.google.com/nihr.ac.uk/pi-standards/the-project/work-in-progress

33 Coad J, Evans R. Reflections on practical approaches to involving children and young people in the data analysis process. *Child Soc* 2007;22:41–52.

34 Mockford C, Murray M, Seers K, *et al*. A SHARED study – the benefits and costs of setting up a health research study involving lay co-researchers and how we overcame the challenges. *Res Involv Engagem* 2016;2:8.

35 Kellett M. Small shoes, big steps! empowering children as active researchers. *Am J Community Psychol* 2010;46:195–203.

36 Franks M. Pockets of participation: revisiting child-centred participation research. *Child Soc* 2011;25:15–25.

37 INVOLVE: Payment and recognition for public involvement. http://www.invo.org.uk/resource-centre/payment-and-recognition-for-public-involvement/

275

EP

Ethics and patient and public involvement with children and young people

Sarah Jane Mitchell, Anne-Marie Slowther, Jane Coad, Shazaan Akhtar, Elizabeth Hyde, Dena Khan and Jeremy Dale

---

Updated information and services can be found at:
http://ep.bmj.com/content/early/2018/02/08/archdischild-2017-313480

These include:

References	This article cites 23 articles, 6 of which you can access for free at: http://ep.bmj.com/content/early/2018/02/08/archdischild-2017-313480 #ref-list-1
Email alerting service	Receive free email alerts when new articles cite this article. Sign up in the box at the top right corner of the online article.

Notes

Achieving beneficial outcomes for children with life-limiting and life-threatening conditions receiving palliative care and their families: A realist review

Sarah Mitchell[1] iD, Karina Bennett[1], Andrew Morris[1], Anne-Marie Slowther[1], Jane Coad[2] and Jeremy Dale[1]

Abstract

Background: Palliative care for children and young people is a growing global health concern with significant resource implications. Improved understanding of *how* palliative care provides benefits is necessary as the number of children with life-limiting and life-threatening conditions rises.

Aim: The aim is to investigate beneficial outcomes in palliative care from the perspective of children and families and the contexts and hidden mechanisms through which these outcomes can be achieved.

Design: This is a systematic realist review following the RAMESES standards. A protocol has been published in PROSPERO (registration no: CRD42018090646).

Data sources: An iterative literature search was conducted over 2 years (2015–2017). Empirical research and systematic reviews about the experiences of children and families in relation to palliative care were included.

Results: Sixty papers were included. Narrative synthesis and realist analysis led to the proposal of context–mechanism–outcome configurations in four conceptual areas: (1) family adaptation, (2) the child's situation, (3) relationships with healthcare professionals and (4) access to palliative care services. The presence of two interdependent contexts, the 'expert' child and family and established relationships with healthcare professionals, triggers mechanisms, including advocacy and affirmation in decision-making, which lead to important outcomes including an ability to place the emphasis of care on lessening suffering. Important child and family outcomes underpin the delivery of palliative care.

Conclusion: Palliative care is a complex, multifactorial intervention. This review provides in-depth understanding into important contexts in which child and family outcomes can be achieved so that they benefit from palliative care and should inform future service development and practice.

Keywords

Child, palliative care, terminal care, healthcare facilities, manpower and services, realist review

---

What is already known about the topic?

- The population of children and young people with life-limiting and life-threatening conditions and associated palliative care needs is rising internationally.
- Specialist paediatric palliative care services provide benefits for children and their families including symptom control and improved quality of life, a feeling of support and achieving a preferred place of care and death, all of which align with current policy.
- Specialist paediatric palliative care services are inconsistent around the world, and their future development has significant resource implications.

---

[1]Warwick Medical School, University of Warwick, Coventry, UK
[2]School of Health Sciences, University of Nottingham, Nottingham, UK

Corresponding author:
Sarah Mitchell, Warwick Medical School, University of Warwick, Gibbet Hill Road, Coventry CV4 7AL, UK.

Appendix 2: Characteristics of studies included in the systematic review

Study	Design & Research Questions	Participants	Intervention	Findings
M. J. Sheetz et al (2012) (98)	What are parents' perceptions about whether a SPPC programme was providing key elements of paediatric palliative care? Are parents satisfied with the service? Questionnaire survey of parents whose children were receiving care from a specific SPPC programme. Salt Lake City, USA	Parents of 65 children who had died while under the care of the programme. CYP with a range of LLCs and LTCs, including cancer and complex chronic conditions, most frequently genetic / congenital, neuromuscular and cardiovascular conditions.	SPPC programme: MDT including pediatrician and nurse practitioner board-certified in PPM, nurse manager, social worker, chaplain. Hospital-based inpatient consultation service and home consultation service after discharge. Age range prenatal-36 years.	65/192 responded (37%). 37% (22) of children had died at home, 53% (32) at the hospital, and 8% (5) at another hospital, 2% (1) in a long term care facility. 21 were also receiving hospice services at the time of death. 95% agreed or strongly agreed that the SPPC team helped them make decisions about their child's care, 5% disagreed. 76% felt the team had helped them set goals for care (others unsure or disagreed), 78% agreed that those goals of care were subsequently met. 13% unsure, 10% disagreed. 95% felt supported in their choices for their child's care. Also 78% felt that physical pain / distressing symptoms were controlled at the end of life, 22% unsure or disagreed. 71% felt symptoms other than pain were controlled adequately. High levels of parent satisfaction with SPPC. SPPC had an important role in education: decision making, the process of death and aspects of the medical system. Feedback included a desire that the team were involved sooner.
L. K. Fraser et al (2013) (100)	What is the impact of SPPCS on the number of hospital admissions in children before their death with cancer? Retrospective cohort study of epidemiological data. Differences in hospital admission patterns were assessed using negative binomial regression Yorkshire, UK	2508 patients aged 0-19 years with cancer from 1990-2009, who were in the catchment area for the SPPCS. .	SPPC based at a children's (aged up to 35 years) hospice with a full time consultant from 2004. 24 hour on call medical service, and a home visiting service.	27.7% of those who had died were referred to SPPC (less than a third). 182 had died and had been referred, 475 had not been referred before they died. No significant difference in terms of demographics. Most commonly referred were those with CNS tumours. Referral to SPPC was associated with a significantly lower rate of planned hospital admissions (IRR=0.60, 95% CI 0.43-0.85). There was no significant difference in emergency (which comprised 97% of admissions) or total hospital admission rates. Children with CNS tumours had significantly less hospital admissions compared to those with leukaemia

				(may be due to the nature of treatment).
G. Groh et al (2013) (101)	Does the involvement of a SPPC team (home care) address the needs of patients and their families and thus lead to an increase in acceptance and effectiveness of SPPC? Prospective non-randomised study with two validated questionnaires; the first during the first week pf paediatric palliative home care (SPPC) service involvement, and the second a few days - six months later depending on clinical condition. Face to face by a trained psychologist. Bavaria, Germany	All primary caregivers of severely ill children receiving SPPC via the PPHC team for the first time between Apr 2011 and June 2012. 40 families. 18 CYP died before study ended. 16 of them at home. Wide range of diagnoses including cancer and complex chronic conditions	Multiprofessional SPPC team consisting of three pediatricians, two nurses, a social worker and a chaplain, all with special training in palliative care. 24/7 on-call.	SPPC was assoc with improved satisfaction with care and quality of care. Significant improvement in children's symptom control and quality of life as perceived by parents. Parents own QoL and burden relief significantly increased and psychological distress and burden decreased. SPPC led to reduced rates of hospitalization and improved caregiver satisfaction with care received including psychological support and support of activities of daily living. Caregiver's felt more informed on disease situation and progression, better taken care of, improved communication with child. Psychosocial support was identified as the most helpful aspect of care. 24/7 on-call service and time for detailed conversations highly valued, as were support with practicalities e.g. ACP, equipment.
L Niswander et al. (2014) (97)	What SPPC are CYP receiving at home, and how is their end-of-life experience perceived by parents and medical personnel. Retrospective cohort review of medical notes of children who died from Dec 2004 – May 2008. New York, USA	36 children who died with a wide range of diagnoses, including cancer, enrolled in the program for a median 1.1 years before they died.	A team consisting of pediatric nurses, pediatric nurse practitioners, a pediatrician board-certified in hospice and palliative medicine, child life specialists, social workers, chaplain, and expressive therapists.	There was a median of 3 hospitalizations (inc planned), 2 emergency room visits in the last 6m of life. Median of 24 home visits (1-121), home visit frequency varied. Symptom control was important, as were "goals of care" discussions, which happened a median of 16 days before death (0-116). 25 had home DNACPR. 15 CYP were aware of impending death (recorded) – tended to be older CYP. 16 children died at home, 20 died in hospital. 16 had recorded PPD. Conclusion was that children who die of complex chronic conditions spend most of their last 6 months at home, community SPPC contributes substantially to their care and comfort.
A. Postier et al. (2014) (99)	What is the healthcare utilization by children prior to enrollment in SPPC	425 Children with a range of diagnoses aged 1-21 under the care of SPPC for at	24/7 access and care co-ordination through home visits by nurses, social workers, child	No of hospital admissions didn't change. Length of stay decreased by two weeks post SPPC involvement,

	compared to the period after enrollment? Retrospective cohort study of electronic medical records, and economic analysis. Minneapolis, USA	least one day between 2000-2010	life specialists, chaplains, music / massage therapists, physicians and volunteers.	with a significant drop in healthcare costs. Largest decreases in resource utilization for the non-malignant group – reduced length of stay in hospital with SPPC involvement, decrease in hospital charges for those under SPPC > 6 months.
A. Herbert et al (2014) (104)	What are the characteristics of the population care for by the SPPC service, what outcomes are the SPPC achieving and how has the service developed? Retrospective cohort review of medical notes of children who were referred to the service in a 2 year period. Brisbane, Australia	150 patients referred over a 24-month period. Wide range of diagnoses.	The SPPC developed from the existing pediatric oncology palliative care service at the RCH, and incrementally expanded over a period of 3 ½ years, commencing with a dedicated pediatrician trained in palliative care, followed by nursing staff and the addition of dedicated allied health staff from 2010. Offers biannual training days and videoconferencing. 24/7 telephone support.	Median duration of contact time with the service was 83 days. Non-oncology diagnosis was likely to result in longer use of the service (>6m). 41% of children died at home and 48% died at hospital. Reasons for referral: Pain and symptom management (29%), Advanced care planning (25%), Community care planning (21%), End of life care (26%).
S. Friedrichsdorf et al. (2015) (96)	How does end-of-life pain and symptom management in children with advanced cancer who received care exclusively from oncology compare with those who received concurrent SPPC home care? What are the differences between the two groups regarding health outcomes inc QoL and location of death. Retrospective cohort survey study of bereaved parents whose children died of cancer Minneapolis, USA	Final sample of 60 bereaved parents of children who died of cancer between 2002-08 at a US tertiary paediatric institution .	PPC nurses, social workers, and chaplaincy. A PPC physician and/or pediatric oncologist or oncology advanced practice registered nurse. 24/7 nursing on-call for home visits	No significant difference in prevalence of symptoms between those referred to SPPC and those who were not. There was a trend towards greater perceived suffering from pain in the group who were not referred to SPPC group. Seizures and nausea/vomiting were the most successfully managed in both groups. More parents in SPPC group wanted their child to die at home and had opportunity to plan this. More children in PPC group did die at home. SPPC children had more fun and more likely to have an experience, which added meaning to their life.

A. Kassam et al. (2015) (105)	Is referral to SPPC associated with improved end-of-life care communication for children with advanced cancer and their families? Questionnaire survey and medical record review examining differences in end-of-life communication for children with advanced cancer who were referred to a SPPC team Toronto, Canada	75 bereaved parents	PACT team Four palliative care physicians, three nurses, two grief support coordinators and one administrative assistant. PACT also draws on the special knowledge of other professionals and volunteers like chaplains, social workers, nurses, bioethicists, physicians, pain experts, volunteers and parents.	Most frequent communication was DNACPR discussion. Least frequent was discussion of death and dying when appropriate, and advice on how to talk to child about this Univariate analysis showed parents more likely to have the following five communication elements if a palliative care team were involved: Discussion of death and dying with parents and with the child, guidance on how to talk to their child, preparing parents for medical aspects of death and sibling support.
K. Widger et al. (2018) (106)	Which children with cancer access SPPC and the impact of accessing SPPC on the risk of experiencing high intensity end of life care during the last 30 days of life Toronto, Canada	572 children who received care through one of five paediatric oncology programs and died	Palliative care (PC) of Specialised Paediatric Palliative Care Services (SPPC)	Results Of the 572 children, 166 (29%) received care from an SPPC team for at least 30 days before death, and 100 (17.5%) received general palliative care. SPPC was associated with a five-fold decrease in odds of intensive care unit admission (OR, 0.2; 95% CI, 0.1 to 0.4), whereas general palliative care had no impact.
S. Nolte-Buchholtz et al (2018) (102)	What are the characteristics of patients referred to nine specialized paediatric palliative home care (SPPHC) teams in Germany, and what are the care goals? Germany	Data on 75 new referrals to SPPHC teams from April 2013 to September 2013.	SPPHC team provision by law in Germany. The law provides quality criteria for the composition of the team, including certified qualification of palliative care physicians	The majority of patients (72%) had non-malignant conditions. Patients were a diverse cohort with complex conditions and use of medical technology including feeding tubes, oxygen, tracheostomy, ventilation and central venous catheters. The provision of care was different for patients with cancer.
P. Chong et al. (2018) (107)	What are the differences in patient outcomes for patients referred to the Paediatric Advance Life Support (PALS) team compared to usual care? What are the differences in	Three year retrospective cohort study (2012-2015)	Star (PALS), a specialist home-based palliative care service supported by a specialist-grade physician	Home-based paediatric palliative care brings improved resource utilization and cost-savings for both patients and healthcare providers, and improves the health-related quality of life of children and their caregivers being able to spend more time at home.

	healthcare expenditure. Quality of life, and caregiver burden? Singapore			
B. Zernikow et al. (2018) (103)	How has end of life care changed in paediatric cancer patients over 10 years? Are changes in the structure of paediatric palliative care associated with changes in the quality of care? Germany	124 families of children with cancer participated in interviews study carried out between 2005 and 2015 at three time points (2005, 2010 and 2015)	SPPHC team provision by law in Germany. The law provides quality criteria for the composition of the team, including certified qualification of palliative care physicians	Parents' perception of symptom occurrence, symptom burden and effectiveness of symptom control remained stable. The availability of paediatric palliative care increased over 10 years, as did quality and satisfaction ratings of care. More children received care at home during the end of life period, and more families had the opportunity to plan the location of death.

Appendix 3: Characteristics of studies included in the realist review

Reference	Year	Country	Type of paper	Aim	Methods	Participants
Wolfe et al Understanding of prognosis among parents of children who died of cancer: Impact of treatment goals and integration of palliative care (141)	2000	USA	Research	To compare the timing of parental understanding that child had no realistic chance for cure with that of the physician.	Interviews with bereaved parents (face-to-face and phone) using questionnaires, and case note reviews; and interviews with oncologists. Statistical analysis.	Parents of 103 children who had died of cancer (one parent per family) and 42 paediatric oncologists
Wolfe et al. Symptoms and suffering at the end of life in children with cancer (115)	2000	USA	Research	To establish whether children with cancer received high-quality end of life care as standard.	Interviews with bereaved parents (face-to-face and phone) using questionnaires, and case note reviews with statistical analysis	Parents of 103 children who had died of cancer (one parent per family)
Contro et al. Family perspectives on the quality of pediatric palliative care (160)	2002	USA	Research	Exploratory study of the experiences of families of paediatric patients at a children's hospital carried out to inform the development of a PPC service	Interviews followed by thematic analysis of qualitative data.	68 family members of 44 deceased children (English and Spanish, mixed diagnoses).
Macdonald et al. Parental perspectives on hospital staff member's acts of kindness and commemoration after a child's death (161)	2005	USA	Research	Study to explore family experience of the hospital memorial service	Interviews with parents of children who had died in ICU, thematic analysis	12 parents of 8 deceased children
Darnill S et al	2006	UK	Parent narrative	To describe a mother's experience of her son's death from cancer	First person narrative	First author is a bereaved mother of a 17 year old son with a brain tumour

The patient's journey: palliative care - a parent's view. (137)						
Steele R et al. Impact on parents when a child has a progressive, life-threatening illness. (156)	2006	Canada	Research	To generate a contextually-grounded description of the experiences of families living with a child who has a neurodegenerative life-threatening illness (NLTI).	Grounded theory study	29 family members from 8 families through observation and interviews
Surkan et al. Perceptions of inadequate health care and feelings of guilt in parents after the death of a child to a malignancy: a population-based long term follow-up (142)	2006	Sweden	Research	To estimate the prevalence of guilt and to identify the care factors and their ability to predict feelings of guilt in parents of children who die from malignancy	Written questionnaire survey study	449 parents who had a child who had died of cancer in Sweden
Hendricks-Ferguson V. Parental perspectives of initial end-of-life care communication. (163)	2007	USA	Research	To examine parents' perspectives of: 1. The timing and method used by healthcare providers to introduce EOL options for their child and 2. What their preference would have been regarding the selected time and method to introduce EOL options	Retrospective descriptive study using qualitative interviews and thematic analysis	28 bereaved parents (19 mothers and 9 fathers). Mixed diagnoses; malignant and non-malignant disease.
Hsiao J, et al Parent and child perspectives on physician communication in	2007	USA	Research	To identify the aspects of physician communication that children with life-limiting illnesses and their parents perceived to be facilitative or obstructive in pediatric palliative care.	Qualitative interview study	20 parent and child pairs of pediatric oncology and cardiology patients (9-21 years) with a poor prognosis

pediatric palliative care. (162)						
Monterosso L, et al Supportive and palliative care needs of families of children with life-threatening illnesses in Western Australia: evidence to guide the development of a palliative care service.(29)	2007	Australia	Research	To elicit the views of parents and service providers to better understand the needs of such families in Western Australia, the extent of service provision currently available to these families and the barriers and facilitating factors associated with supportive and palliative care.	Phase 1 consisted of a series of surveys and Phase 2 consisted of interviews with parents and service providers.	Survey of 129 parents; a cancer group (n = 19, all bereaved) and non-cancer group (n = 110) Interviews with 38 parents; cancer (n=10) and non-cancer (n=28) and 20 service providers
Konrad Cohen S et al. Mothers' perspectives on qualities of care in their relationships with health care professionals: The influence of relational and communicative competencies. (164)	2009	USA	Research	To explore mothers' perspectives on helpful and unhelpful qualities of health care provision.	Psychological phenomenological study with open interviews and comparative analysis as per Paavilainen and Astedt-Kurki (1997)	12 mothers who had primary responsibility for the day-to-day care of seriously ill and dying children
Hechler T et al. Parents' perspective on symptoms, quality of life, characteristics of death and end-of-life decisions for	2008	Germany	Research	To investigate bereaved parents' perspective on 1) symptoms and quality of life, 2) characteristics of the child's death, 3) anticipation of their child's death and care delivery, 4) end-of-life decisions and	Semi-structured interviews	Parents of 48 children who had died of cancer (31 boys, 17 girls)

children dying from cancer. (143)				5) impact of the child's death on the parents and perceived social support by the health care team.		
Dighe M et al Parental concerns in children requiring palliative care (144)	2008	India	Research	To identify the concerns of parents of children with advance incurable cancers, and to elicit their attitudes toward revealing the diagnosis and prognosis to the sick child	Qualitative semi-structured interview study. Thematic analysis	31 parents of 20 children (mothers n=5, fathers n=4, both =11)
Monterosso et al Supportive and palliative care needs of families of children who die from cancer: an Australian study Australia (145)	2008	Australia	Research	To elicit parents understanding of palliative care, their experiences of palliative and supportive care received during their child's illness, and their palliative and supportive care needs.	Qualitative semi-structured interview study	24 parents of children who died of cancer
Monterosso et al The supportive and palliative care needs of Australian families of children who die from cancer (146)	2009	Australia	Research	To identify the perceptions of parents of children who died from cancer regarding the palliative and supportive care they received in hospital and in community settings.	Face-to-face or telephone questionnaires (quantitative)	69 bereaved parents of children who died from cancer from 3 Australian states
Zelcer et al Palliative Care of Children with Brain Tumours: A Parental Perspective (147)	2010	UK	Research	To explore the end of life experiences of children with brain tumours and their families	Semistructured focus group interviews with parents of children who had died of brain tumours	25 bereaved parents of 17 children, in three focus groups
Knapp C et al. Family Support in Pediatric Palliative Care: How Are Families Impacted by their Children's Illnesses? (168)	2010	USA	Research	To determine how families are impacted and what factors are associated with greater impact.	Cross-sectional telephone survey using the Impact on Family (IoF) scale, multivariate analysis	85 parents whose children had life-limiting illnesses and were enrolled in a publicly funded hospice-based pediatric palliative care program.
Neil L et al. 2010 Learning to live with	2010	Ireland	Literature review	An analysis of literature on paternal and maternal development and	Systematic literature review (2 papers)	2 papers

childhood cancer: a literature review of the parental perspective. (132)				resilience through the experience of having a child diagnosed, treated, and possibly die from cancer.		
Contro N et al. Away from home: Experiences of Mexican American families in pediatric palliative care (166)	2010	USA	Research	To describe the experiences of Mexican American family members who immigrated to the United States and then experienced the death of a child	Semi-structured interviews and thematic analysis	38 bereaved participants representing 21 families of Mexican American descent
Menezes A. Moments of realization: life-limiting illness in childhood-perspectives of children, young people and families. (167)	2010	UK	Research	To understand the perceptions and experiences of the child, and to frame that understanding with family insights.	Grounded theory study. Case studies using participant observation and interactive methods drawn from education research.	11 children and young people affected by life-limiting conditions. Parents and siblings were also vital contributors (39 participants from 10 families).
Miedema B et al. 'You can only take so much, and it took everything out of me': Coping strategies used by parents of children with cancer. (148)	2010	Canada	Research	To assess the coping strategies of parents of children with cancer	Semi-structured interviews. Thematic analysis with a focus on parental coping strategies.	28 French and English families who had had a child diagnosed with cancer in the last ten years.
Knapp C et al. Factors affecting decisional conflict for	2010	USA	Research	To examine factors affecting decisional conflict for parents	Telephone survey to establish decisional conflict scores Statistical analysis	140 parents of children with life-limiting illnesses enrolled in Florida's publicly funded paediatric palliative care programme (Partners in Care: Together for Kids).

parents with children enrolled in a paediatric palliative care programme. (165)						
Fauman K et al. Predictors of depressive symptoms in parents of chronically ill children admitted to the pediatric intensive care unit. (169)	2011	USA	Research	To identify factors in the pediatric intensive care unit (PICU) patient population that may result in increased risk of depressive symptoms in their parents	Six-month, prospective, observational study in a tertiary-level PICU on parents of chronically ill children admitted to PICU. Parents were assessed by background questionnaire and standardized depression scale.	61 parents of 39 children (mothers n=37, fathers n=24). 18 were admitted to PICU electively after surgery
Tomlinson D et al. Concordance between couples reporting their child's quality of life and their decision making in pediatric oncology palliative care. (149)	2011	Canada	Research	To (1) describe concordance between fathers' and mothers' evaluation of quality of life (QoL) and (2) determine parental correlation for how factors such as hope, anticipated QoL, and prolonged survival time influence decisions between supportive care alone versus aggressive chemotherapy.	Cross-sectional interview survey study using scales and tools. Intraclass Correlation Coefficient	Both parents of 13 children with cancer and no reasonable hope of cure
Kirk S, et al An exploration of parents' and young people's perspectives of hospice support. (170)	2012	UK	Research	To investigate parents' and young people's perceptions of hospice support and identify how support could be improved	A mixed-method approach was used involving a postal survey of families and in-depth qualitative interviews with a purposively sampled subsample of parents and young people.	108 questionnaires returned (71 parents and 37 bereaved parents) Interviews with 12 parents and 7 young people
Price J et al. Comparing the needs of families of children dying from malignant	2012	UK	Research	To examine the experiences of bereaved parents concerning the care of children with cancer,	Qualitative interview study with bereaved parents, two centres (hospital and hospice) Thematic analysis	25 parents of 16 children (cancer n=6, non-cancer n=10)

and non-malignant disease: an in-depth qualitative study (27)				compared to those who died from a non-malignant condition		
Robert R, et al. Bereaved parents' perspectives on pediatric palliative care. (171)	2012	USA	Research	Focus groups with parents whose children were age 10 years and older at the time of death.	Qualitative focus group Thematic analysis	14 parents from 9 families (36%) agreed to participate in the study. Three focus groups were conducted, with two, seven, and five parents in the first, second, and third focus groups, respectively.
Von Lutzau et al 2012. Children dying from cancer: parents perspectives on symptoms, quality of life, characteristics of death and end-of-life care decisions (150)	2012	Germany	Research	To investigate the experience of children who died of cancer.	Qualitative interviews of bereaved parents of children with cancer	Bereaved parents of 48 children (?) known to a palliative care service
Heinze et al Parental decision making for children with cancer at the end of life (133)	2012	USA	Research	To examine the current research on parental decision making at the end of life and to elicit clinical implications	Meta-ethnography (systematic review)	10 studies, all descriptive (7 qualitative, 3 quantitative)
Gupta V, Prescott H. "That must be so hard"- Examining the impact of children's palliative care services on the psychological well-	2013	UK	Research	To measure parental stress and the psychological wellbeing of parents of newly referred children with life-limiting and life-threatening conditions to a palliative care service. Measures were administered at the point of referral and at 12 months follow-up.	Questionnaire survey study	20 complete sets of data

being of parents. (172)						
Gilmer M, et al. Parental perceptions of care of children at end of life. (173)	2013	USA	Research	To describe parental perceptions of the care of hospitalized, terminally ill children in the areas of (1) clinical management, (2) interdisciplinary support, and (3) pain and symptom management.	Mixed methods study using qualitative and quantitative (questionnaire) telephone interviews	15 bereaved parents of children who had died in hospital
Gaab et al 2013 Primary Caregivers Decisions Around Communicating About Death with children involved in pediatric palliative care. (174)	2013	New Zealand	Research	To investigate why parents in NZ speak or avoid speaking with children about the end of life	"Primary caregivers" (included extended family members) Inductive thematic analysis	19 primary caregivers from 11 families. Children had cancer (n=10), heart conditions (n=5) muscular dystrophy (n=3) and other illness (n=1) and were known to PPC services
Brooten D, et al Parent's perceptions of health care providers actions around child ICU death: What helped, what did not. (51)	2013	USA	Research	To describe parents' perspectives of health care provider actions that helped or did not around the time of infant/child's intensive care unit (ICU) death	Semistructured interviews Thematic analysis	63 parents (Black, White, and Hispanic) 7 months post infant/child death,
O'Shea E, Kanarek R. Understanding pediatric palliative care: What it is and what it should be. (139)	2013	USA	Case study	The case study presented here tells the story of David Karanek, and his family. David was the son of coauthor Robin Kanarek, BSN, RN, and her husband, Joe. Details of David's journey, and his family's, from the point of diagnosis illustrate how health care providers might benefit from improved education in pediatric palliative and end-of-life care	First person narrative	Mother of a son who died from leukaemia
Rempel GR et al Parenting under	2013	Canada	Research	To describe the process of parenting young children who have	Grounded theory study	53 interviews with 25 parents (15 mothers, 10 fathers) and 28 grandparents (17 grandmothers, 11

pressure: a grounded theory of parenting young children with life-threatening congenital heart disease. (157)				survived hypoplastic left heart syndrome to inform parent-focused interventions.	Analysed using open and focused coding, constant comparative analysis and memoing	grandfathers) of 15 young children (6 months–4Æ5 years) who had undergone the Sano surgical approach for hypoplastic left heart syndrome.
Steele A et al Bereaved parents and siblings offer advice to healthcare providers and researchers.(151)	2013	USA & Canada	Research	To determine how to improve care for families by obtaining their advice to healthcare providers after a child's death from cancer	Qualitative interviews with thematic analysis	99 participants (36 mothers, 24 fathers, 39 siblings from 40 families)
Sheetz M, et al. Parents' perceptions of a pediatric palliative program. (98)	2013	USA	Research	To assess parents' perceptions about whether a pediatric palliative care program was providing key elements of pediatric palliative care as described in the literature and to assess parental satisfaction with services.	Written questionnaire survey	65 parents whose children died while receiving services from a palliative care service
Wang J et al. The experience of Chinese American parents of children with life-limiting illness: A comprehensive review. (134)	2013	USA	Literature Review	To describe Chinese American parents' experiences during their children's end-of-life period from a culturally informed perspective	Literature review	6 articles included
Steele R et al. Charting the territory: symptoms and	2014	Canada & USA	Research	To describe the symptoms associated with progressive, non-	Observational longitudinal study	275 children from 258 families

functional assessment in children with progressive non-curable conditions (158)				curable, genetic, metabolic or neurological conditions		
Blume et al Parental Perspectives on Suffering and Quality of Life at End-of-Life in Children With Advanced Heart Disease: An Exploratory Study (159)	2014	USA	Research	To describe parent perspectives regarding the end-of-life experience of children with advanced heart disease.	Written questionnaire survey	50 bereaved parents
Sullivan J et al. What parents want from doctors in end-of-life decision-making for children. (176)	2014	Australia	Research	To examine parents' views and experiences of end-of-life decision-making.	Semistructured interviews	25 bereaved parents
Gaab et al Siblings caring for and about pediatric palliative care patients (171)	2014	New Zealand	Research	To identify the concerns of siblings of pediatric palliative care (PPC) patients.	Semistructured interviews Qualitative inductive thematic analysis.	8 siblings of PPC patients aged 9 to 22
van der Geest et al Parents' Experiences of Pediatric Palliative Care and the Impact on Long-Term Parental Grief (179)	2014	Holland	Research	To explore parents' perceptions of the interaction with health care professionals (communication, continuity of care, and parental involvement) and symptom management during the pediatric palliative phase, and to investigate	Retrospective cross-sectional study using a set of questionnaires Quantitative analysis	89 parents of 57 children who died of cancer

				the influence on long-term grief in parents who lost a child to cancer.		
Melin-Johansson et al When a Child Dies: Parents' Experiences of Palliative Care - An Integrative Literature Review. (135)	2014	Sweden	Literature review	To contribute to the knowledge and understanding of parents' experiences of palliative care when a child in the family is dying or has died due to illness.	Integrative literature review	Nine articles were included
Rodriguez A, King N. Sharing the care: the key-working experiences of professionals and the parents of life-limited children (180)	2014	UK	Research	To investigate the roles and care experiences of professionals working in paediatric palliative care To explore the lived experiences of parents of children with LLCs To highlight where these perspectives do or do not converge.	Interpretive/hermeneutic phenomenological research (focus groups and interviews with professionals and oarents)	Three focus groups with 21 professionals (n=11, 5 and 5) working with children with LLCs (Table 1). In addition, 20 individual semi-structured interviews were conducted with the parents (18 mothers and 2 fathers) of children diagnosed with a LLC.
Hill D et al. Problems and hopes perceived by mothers, fathers and physicians of children receiving palliative care. (175)	2015	USA	Research	To describe the problems and hopes reported by mothers, fathers and physicians, examine concordance, parental perceived agreement and how the problems identified by parents were addressed.	Semi-structured interviews with specific questions about problem and hope categories and domains. Paired t-tests to compare parent and physician responses across these domains.	71 parents and 43 clinicians (those considered to be most involved in the child's care)
Coad et al Exploring the met and unmet needs of life-limited children, young people and families (33)	2015	UK	Research	To describe the met and unmet needs of children and young people up to the age of 25 years with long term life-limiting and life-threatening conditions	Qualitative interviews and a focus group using Appreciative Inquiry. Framework analysis	59 adults and 18 children and young people

Gans D et al Impact of a pediatric palliative care program on the caregiver experience. (181)	2015	USA	Research	To assess the impact of a community based paediatric palliative care program on parents / caregivers levels of stress and worry	Survey study conducted at intervals with parents of children under the care of the program	93 caregivers (first interview) 50 (second survey) 18 (third survey wave). Ethnically diverse population
Kuan GL et al. Parents' perspectives on the important aspects of care in children dying from life limiting conditions: A qualitative study. (182)	2015	Malaysia	Research	Malaysia has yet to develop a national PPC policy. In anticipation of this, as part of a needs based qualitative study, parents' views were solicited, as to the unmet needs of their children during the terminal phases of their illness.	Semi-structured interiews Thematic analysis	15 parents of nine deceased children (range of diagnoses)
Kars et al The Parents Ability to Attend to the "Voice of their Child" with incurable cancer during the palliative phase (152)	2015	Holland	Research	To describe and explain parents actions and handling of the "voice of the child"	Qualitative interviews and thematic analysis	34 parents of 17 children with incurable cancer
van der Geest et al Parents faith and hope during the paediatric palliative phase and the association with long term parental adjustment (178)	2015	Holland	Research	To explore the role of faith and hope as a source of coping and indicator of long-term parental adjustment.	Questionnaire study to explore faith, hope and sources of coping	89 parents of 57 children who died of cancer

Bjork M et al. Like being covered in a wet and dark blanket - Parents' lived experiences of losing a child to cancer. (153)	2016	Sweden	Research	To illuminate parents' lived experiences of losing a child to cancer.	Interviews and a narrative about parents' experiences of losing a child to cancer were gathered from parents whom had participated in a longitudinal study across the child's illness trajectory. The analysis of the data was inspired by van Manen's hermeneutic phenomenological approach.	Six parents
Jalmsell L Children with cancer share their views: Tell the truth but leave room for hope. (154)	2016	Sweden	Research	One in five children diagnosed with cancer will die from the disease. The aim of the study was to explore how children with cancer want to receive bad news about their disease, such as when no more treatment options are available.	Qualitative interviews Thematic analysis	10 children with cancer aged 7-17 years
Collins A, et al. Lived experiences of parents caring for a child with a life-limiting condition in Australia: A qualitative study.(28)	2016	Australia	Research	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.	Cross-sectional, prospective, qualitative study. Transcripts were subjected to a thematic analysis, underpinned by an interpretative phenomenological framework.	14 parents recruited form a statewide paediatric hospice
Zimmermann K et al. When parents face the death of their child: a nationwide cross-sectional survey of parental perspectives on their	2016	Switzerland	Research	To assess the EOL care perspectives of a Swiss population-based sample of bereaved parents who had lost a child due to a cardiac, neurological or oncological condition, or during the neonatal period in the years 2011 or 2012.	Questionnaire survey	135 families

child's end-of life care. (183)						
Montgomery K, et al. Experiences of pediatric oncology patients and their parents at end of life: A systematic review. (136)	2016	USA	Research	To describe the experience of pediatric oncology patients and their parents during end of life, and identify gaps to be addressed with interventions.	Systematic review	43 articles
Nelson M, et al 2017. 'Just gripping my heart and squeezing': Naming and explaining the emotional experience of receiving bad news in the paediatric oncology setting. (155)	2017	UK	Research	To explore recipients' perspectives on the range and origins of their emotional experiences during their 'bad news' consultations.	Data was collected using emotional touchpoint storytelling. The names (descriptors) given to the emotional experiences were linguistically classified. Explanations of their perceived origins were examined using applied thematic analysis.	Four bereaved families of children who had changed from active treatment to palliative care in paediatric oncology.
Widger et al. Predictors of Specialized Pediatric Palliative Care Involvement and Impact on Patterns of End-of-Life Care in Children with Cancer (106)	2017	Canada	Research	To determine which children with cancer access SPPC and the impact of SPPC on the risk of experiencing high-intensity end of life care	Retrospective cohort population study using cancer registry data	572 children who died

Nimmo S 2018 Letting my daughter go (138)	2018	UK	Parent narrative	"What your patient is thinking" article	First person narrative	One parent
Kavas M How to increase the quality of suffering experience: lessons derived from the diary narratives of a dying adolescent girl (140)	2018	Turkey	Research	To provide insight into the lived experience of dying	Hermeneutic analysis of a personal diary	15 year old who died of advanced cancer

Appendix 4: Realist review: Supporting evidence

	Contexts	Mechanisms	Outcomes
Family adaptation	**Child develops a life-limiting or life-threatening condition** "the ubiquitous sense of apprehension and uncertainty felt by parents throughout their child's cancer illness trajectory, regardless of their determination to 'fight' cancer"."Some parents described their response to the cancer diagnosis as shock and disbelief, and immediately attempted to find a rational explanation for the unexplainable and the unknown. Other parents responded with stoic resistance and explained the way in which they adapted to their child's illness and care requirements on dayto-day basis while, with what appeared to be an automatic suspension of their own personal life interests. Some parents experienced a range of emotions in a short space of time as they attempted to assimilate the idea that their child had cancer. For many parents there was a sense of hope juxtaposed with uncertainty and apprehension, while trying to live from day-to-day with the practical realities of treatment and care. Monterosso et al 2008 "the parents in this study had little time to adjust to the loss of their envisioned perfect baby as they were	**Family adaptation to a situation that is against cultural norms, framing and re-framing hopes and expectations** "no parent wants to be told that their child is at risk of dying" "negative psychosocial impacts can be associated with the diagnosis of a life-limiting illness. Studies have shown that parents grieve for their children's loss of health along their child's continuum of illness, although the intensity of the parent's grief varies" "55% of parents indicated that no one understands their burdens" Knapp et al 2010 "Parental coping mechanisms included striving to maintain normality and finding spiritual strength through maintaining hope and in the resilience of their child" Zelcer et al 2010 "The findings of the current study may reflect a similar phenomenon of adaptation in depressive symptoms for these parents. Many experts in palliative care and quality of life also speak of a change over time in ''meaning finding'' for terminally ill patients who lose physical abilities over time. This may also occur for caregivers of chronically ill children, thus allowing	**Vulnerable family** "As a result, family structure and organization become permanently altered" Monterosso 2008. "Parents often needed help to alleviate the impact of their experience, but it was not always available in a useful manner" Steele R 2006. "The commonest parental reaction was a feeling of sadness and was expressed by all of those interviewed. 90% expressed helplessness." Dighe et al 2008 "Knowing that your child is going to die imposes complicated and profound burdens on parents and families. No parent is prepared to care for a seriously ill child and no parent is ever prepared for a child's death; to contemplate the death of one's child is to imagine the unthinkable. For parents, feelings of loss and grief begin at the time of diagnosis and continue throughout the course of children's lives cycling

immersed in caring for a seriously ill baby" Rempel et al 2013	them to change their expectations and adapt to circumstances rather than developing increasing caregiver fatigue and sorrow, which could otherwise lead to depressive symptoms" Fauman K et al 2011	through periods of hopefulness, despair, normalcy, and crisis" Konrad Cohen et al 2008
"Immediately after receiving the diagnosis, parents commonly felt lost and full of despair about their child's future. However, they also had hopes and expectations that they would receive all the care and support needed. The reality of what services were available and the level of help, care, and support they could draw on was a shock." Rodriguez et al 2014	"Growing increasingly attached was associated with the subprocesses of seeing the baby as their child and imagining the future for their child. The tension in this second phase related to parents' desire to physically and emotionally nurture their baby while wanting to protect themselves in case their baby did not survive." Rempel et 2013	"65% or more of parents agree that their child's illness results in parental fatigue (71%), giving up things (71%), having to change plans at the last minute (76%), difficulties finding a reliable caregiver (77%), and that living with an ill child can sometimes feel like a rollercoaster ride (90%)" Knapp C et al 2010
"Realizing and adjusting to the inconceivable was associated with two sub-processes: realizing the precariousness of survival and adjusting expectations. It was inconceivable that their infant had a potentially lethal condition as parents and grandparents alike had expected a healthy child; not realizing 'that things went wrong these days'. (GM1) A mother of a postnatally diagnosed baby described feeling 'more scared than anything' and devastated that her baby was 'dying of course'. (M2) As parents acknowledged how misaligned the potential for death was with their expectations of a healthy baby, they simultaneously began to adjust expectations" Rempel et al 2013	"A third phase of Parenting under Pressure required parents to recognize potential problems and make critical decisions in response to changing conditions and new crises." "A father said, 'You're always hoping for the best but have in your mind that things may actually not work out that well', (F16) and a mother stated: 'you keep your guard up'".Rempel et al 2013	"Appraisal-focused coping strategies involved trying to stay "positive" and "making positive comparisons." Problem-focused coping involved behaviors such as being an advocate for the child and seeking information. The majority of parents, however, described using emotion-focused coping behaviors such as trying to avoid "feeling too much" by hiding difficult emotions and "escaping" from problems. Others used more positive emotion-focused coping behaviors such as humor, seeking support (informal or formal), or writing diaries. A small group of parents used ineffective coping strategies (alcohol abuse, misdirected anger) that added to family stress." Miedema B et al 2010
"Parents in the cancer group reported that health professionals had kept them well informed about their child's condition and treatment plan. Conversely, parents in the non-cancer group felt physicians' discussions regarding their child's diagnosis and		"Trapped inside the house. The experience of feeling 'trapped inside the house' describes parents' physical and social isolation from community, their exclusion from the workforce and the associated impacts on their health and well-being that resulted from

treatment was fragmented, with some parents stating their approach was 'too considerate' and the severity of their child's illness often de-emphasized."Monterosso et al. 2007

There are a series of significant fluctuations in the child's condition

"All parents talked about the uncertainty that characterised their day to day living throughout their child's illness. Uncertainty was described by parents as fluctuating, according to, for example, periods or remission or relative stability. However the focus of uncertainty differed. Parents of children with cancer recounted an oscillation between hope for recovery, fear that cure was impossible and eventual knowledge of impending death. Although some parents of children with non-malignant conditions discussed hope for cure, their uncertainty focussed not on if their child would die, but when or how death would occur" Price et al 2012

"The parents' ability to come to terms with a potentially devastating set of circumstances in a relatively short time period was further evident in their accounts of their baby's first surgery. 'A lot of the pressure came off' (GF5) after the first surgery related to the baby's 'miraculous' (M6, M7) survival, and the parents and grandparents recalled that they 'just felt relieved' and 'could all kind of function seminormally' (GF5) for the remainder of that initial hospitalization, until the next surgery. Ongoing survival fuelled parental hope and worry as did the

accepting the role of primary caregiver" "Seclusion from community. Parents described their seclusion from the broader community, owing to their role as primary caregiver, as one of the most difficult aspects of their experience. Life was contained to the home where routines, equipment and care supports were established, facilitating ease and comfort for the child" Collins et al 2016

Expert family

"Family coping strategies included siblings, parents and carers educating themselves and knowing all they need to know to help them to deal with what was happening" Coad et al 2015

"Not only did parents have to adjust expectations about their sick baby but they also had to adjust their expectations concerning their family given the new demands related to a sick child" Rempel et al 2013

	inconceivability of their child dying and their growing attachment to their baby" Rempel et al 2013 This study identified several 'moments of realization', representing times in the child's life when participants recognized the real threat to the child's life, these included: questions of inheritance, diagnosis and prognosis, acute loss of abilities, slow deterioration and life-threatening surgery" Menezes 2010 **Sources of support** "Families may become closer or they may become more spiritual and hopeful. Family friends, school mates, teachers, and the community may rally around a child who is ill and hold charity events or start foundations." Knapp C et al. 2010 Studies of parent-to-parent support suggest that only parents and not professionals can provide mentorship to other parents (Singer et al., 1999). This perspective is based on the premise that at the heart of collective understanding is ''perceived sameness'' that heightens the connection between parents. Konrad Cohen S et al. 2009	

The child's situation	Child has own interests and priorities	Child becomes ambivalent towards healthcare discussions	Child is passive recipient of care
	"Three important aspects were found with regard to how the children wished to receive bad news. They wanted to receive honest and straightforward information while still being allowed to maintain hope, they wanted to receive information at the same time as their parents, and they wanted to receive information in a way that could be understood by a child of their age". Jalmsell et al 2016	"Parents described the child's ambivalence to talk about death and the importance of the child having control regarding end-of-life discussions: "Our daughter wanted to talk about [terminal cancer], then didn't … . [A doctor asked her], 'What are you afraid of? … Dying? … Why?' … That made it easier for her to talk to us, … to be in control … . She could plan her funeral." Robert et al 2012	Parents demonstrated a kind of resilience that often mirrored the same kind of resilience observed in their children. Monterosso et al 2008
	"The children agreed that there was no good way to deliver bad news." Jalmsell et al 2017	"Moreover, children want to be heard. Studies have overwhelmingly found that children, especially adolescents, want to participate in making medical decisions.[3,4] Children also want to please their parents, which may cause conflict if their treatment preferences differ." Knapp et al 2011	"noteworthy was the observation that many of these same parents were unable to acknowledge or discuss their child's terminal state in the child's presence, even though they knew tacitly that their child was aware of this end point" Monterosso 2008.
	"Children and their families were living as normal a life as possible in abnormal circumstances" Coad et al 2014		"About half of the caregivers felt that the conversation was not necessary because the child already understood all the necessary information" "Many caregivers were aware that patients and siblings knew the reality of the situation … caregivers who spoke about death or prognosis with their children acknowledged their children's expertise and knowledge in their own status." Gaab et al 2013
	"her illness pushed her further away from life" Volkan Kavas 2018	**Parents fear "getting it wrong"** "I didn't know then, and I still don't know the right way—if there is one. What I did impress on Andrew, on his sisters, and on myself was acceptance. Don't fight it. Everything that could be done medically had been done. We fought for life as hard as we could, now we will accept what comes next. I think this worked to a limited extent—certainly Andrew faced his last weeks with serenity, dignity, and forbearance	"During the entire course of David's illness and treatment, his father and I were always aware that he could lose his life. At the age of 11 and with no knowledge or experience of cancer, David didn't fully grasp the seriousness of his illness; in fact, he was relieved when he learned his diagnosis. Finally, there was a name for the cause of his problems. When David's cancer recurred at age 15, his understanding was significantly more advanced. By this time he had
	"Young people wanted to visit the hospice more frequently not only because they enjoyed the activities but also because visits were often their only contact with close friends." It is a break and you get to see each other. With our short life expectancy we don't see each other enough. So they are really		

precious to me. (Young Person Interview 05)" Kirk S et al 2012

The importance of the children continuing to "live their lives" despite the hardships they were enduring. Schooling in particular was mentioned frequently, and the ability to complete a grade or simply to continue attending was referred to with a sense of pride and accomplishment. The maintenance of childhood friendships and the involvement and support of peers was of significance" Zelcer et al. 2010

"Interestingly, these children had longer time to consider this question and they might have met other children who received bad news, or even died, and may be realised that they might one day be in that situation themselves. Therefore, it is reassuring that these three children communicated the same message as the other children." Jalmsell et al 2017

"Parents sometimes stated that they experienced that the child wanted to protect them from sadness and to keep their spirit up, not giving up hope. Some parents experienced that their child searched for their "permission" to die. It was like she had to get our permission to die. Then she should not feel that it is a failure on her part, its ok." Bjork et al 2017

which also typified his behaviour throughout his illness." Darnhill S et al 2006

"Primary caregivers avoided talking about the child's mortality with their child patients and their siblings to protect them" "The caregivers reasons are … preventing negative reactions, losing hope, and blame or regret." Gaab et al. 2013

"Parents to a child with a severe malignant disease experience it as difficult and demanding to talk with their child about his/her imminent death. Kreicbergs et al. (2004) found that parents who have lost a child to cancer, who sensed that their child was aware of their upcoming death, in hindsight regretted not talking to the child about it." Bjork et al 2016

learned about all the medications and their side effects. Devastated that he was going to have to undergo more treatments, he asked, "Why me? What did I do wrong?" I felt at a loss for words. I wasn't prepared to answer this question." O'Shea 2013

"While interviewers asked whether the child was told she or he was dying, results were not included in this analysis, because most children in this sample were infants, so they were not informed about how sick they were. The 14 year-old and 3-year-old patients represented had advanced brain tumors, and the 8-year-old patient endured a cardiac arrest related to keto-acidosis; thus, these children were comatose during their last days" Gilmer et al 2013

Parents and child develop a tacit understanding that death is possible

"Mirac stated through her entire diary narrative that she felt like she "saddened her family, especially her mother, for no reason and because a burdern to them" Volkan Kavas 2018

"The first theme in the participants' explanations of the origins of their experiences centered on the perception of awareness, in terms of both the internal process of becoming aware, and the external process of being made aware" Nelson et al 2017.

Five of the nine children were aged 10-14years, yet in only one family was communication open, with the patient fearing he will be forgotten should his toys be given away. "Mum, please forgive me for always taking it out on you." (voice breaks and cries), Mum, when I am gone, please do not give my toys away." Kuan et al. 2015

Parents become surrogate decision makers

"In children, communication about end-of-life issues may be more challenging because the parent is almost always the surrogate decision maker" Wolfe et al. 2000

"Paediatric patients did not have adequate opportunity to interact with any health professionals on their own." "The major barrier was the attitude of parents who did not permit the palliative care team to interact alone with the child." Dighe et al 2008

"The interviews showed that parents often represented their child's voice. This was, for example in presenting the child's suffering to the general practitioner, or discussing the child's quality of life with the oncologist, to decide on further options for treatment This seems an almost natural act. None of the parents questioned his or her ability to represent their child's voice" "parents use direct and indirect

	strategies to gain insight into their child's inner perspective" Kars et al 2015 "All parents spontaneously gave vivid descriptions of how their child acted and coped with situations with which he or she was confronted. The parents' stories were interspersed with comments reflecting their view of their child, such as their pride about their child's coping with a new setback or how their child managed the suffering of symptoms." Kars et al 2015		
Relationships with healthcare professionals	**Importance of continuity of care** "Parents noted that different health professionals could ask the same questions several times, which was disturbing" Melin-Johansson et al. 2014 "All parents indicated that they were satisfied or very satisfied with the availability of health care providers [in hospital]" Gilmer et al 2013 **Individual professional's motivation / characteristics** "Both children and parents identified five domains of physician communication deemed to be highly salient and influential in quality of care. These included relationship building, demonstration of effort and	**Respect for the family circumstances and Advocacy** "A majority of both children and parents endorsed physician relationship-building skills, skills that facilitated the growth of trust, credibility, rapport, and friendship, reporting that these traits increased their comfort level when speaking with a physician. Children and parents appreciated doctors who took the time to get to know the patients as individuals and develop a friendship with the patients. Hsaio et al 2007 "The family needs considered most important were as follows: to know questions could be asked at any time, to feel that the health care professionals were sincere in caring about their child, to know what treatment their child was receiving, to know when to expect side effects to occur and to have trust in the	**Trusted authentic relationships** "Relationships between the healthcare team and parents were identified as critical as they enabled clear and honest communication. Relationships can be developed through compassion and small acts of kindness. These relationships can have a therapeutic effect and can contribute positively to the memories of the family" Monterosso et al 2007 "Parents consider genuine communication with sincere and honest provision of information to be most central" Zimmerman et al 2016 **Shared emotional impact**

competence, information exchange, availability, and appropriate level of child and parent involvement. Parents identified coordination of care as another important communication domain" Hsaio et al. 2007

"Parents maintained that professional carers who provided end-of-life care to their children require specific paediatric knowledge and experience. The parents in the cancer group stated they felt 'extra stamina' especially during their child's terminal phase and felt naturally inclined to be the carer of their child. Parents from the cancer group who used the services of community-based medical and nursing staff during the end-of-life phase of their child cited a lack of familiarity with the management of their child's medication (especially pain relief) and nutrition. Some parents of children with non-cancer diagnoses were concerned about the skills of the carers and reported that in-home respite or hospice may be useful as long as there was a long period of familiarization prior to use" Monterosso et al 2007 .

"One finding that differentiated the accounts of these mothers from those of parents of children with chronic health conditions and disabilities was the mentorship and support they felt from professionals who identified as having been trained in palliative and end-of-life (EOL) care. Mothers were not specifically asked about the educational backgrounds of the professionals they encountered; however, it was noteworthy how many of them named EOL training as

health care system". "Parents consistently referred to the importance of the quality of interpersonal relationships between them and health care professionals, and between their children and health care professionals. Further, parents clearly articulated the need for honest, open, authentic, and therapeutic relationships as an imperative to facilitate quality care of the dying child to ensure a 'good death'". Monterosso 2009

"It was notable that parents only felt able to have this break because of the trust and confidence they had in the staff to care for their child. They saw their child as being in 'safe hands' in the hospice" "hospice staff not only had the knowledge and skills to care for their child but also the relationships they had developed with their children and their individualized knowledge of them." Kirk S et al 2012

"what helped most were compassionate, sensitive, caring staff; understandable explanations regarding infant's/child's condition; experienced, competent nurses; perceptions that providers did everything to help the infant/child; and parents' involvement in care and decisions" Brooten et al 2013

"emotional care and communication generated the most discussion. The number of related comments for these two themes was 5 to 6 times than the number of comments regarding the content areas of decision

"Paradoxically, although medical knowledge and technical savvy are continuously being improved and refined, both patients and health providers continue to voice dissatisfaction with the state of mainstream medicine and the adjunctive loss of bedside manner " Konrad Cohen et al 2008

a component of their experience of professional competency" Konrad Cohen et al 2009

"Family members offered advice to healthcare professionals about their need to feel supported and cared for throughout the child's illness. A father encouraged healthcare providers to "realize that (these) are special people that ya'll work with." A mother added, "These kids are dying, and they know they are dying. Some of them (healthcare providers) need to be more compassionate." Steele et al 2013

Since 75% of children with cancer will be cured of their disease, paediatric oncologists have comparatively less experience communicating about terminal prognoses and may find it difficult to do so. This hypothesis is supported by the finding that although nearly all parents reported having a discussion at some point with a medical caregiver about their child having no realistic chance of cure, only 49% of parent reported that they came to understand that their child was terminally ill through this discussion" Wolfe et al 2000

Negative experiences

"A single event could cause parents profound and lasting emotional distress. Parents recounted incidents that included insensitive delivery of bad news, feeling dismissed or patronised, perceived

making, spiritual care, or symptom management" Robert R et al 2012

Ability to bear witness to the child and family situation (or not)

"Sometimes caregivers avoided the topic to protect relationships between individuals involved with their families, perhaps in an effort to protect social norms".Gaab et al 2013

"We knew how serious it was all along, but we had the intention of keeping positive. The one bitterness I have left … for some reason, some doctors feel that you're not allowed to have hope, that you're just supposed to accept it and move on." Contro N et al. 2002

"Breaking bad news is perceived by practitioners as one of the more challenging aspects of their work, even by those practitioners who find themselves in this situation on a relatively regular basis [1,2]. Clinicians may exhibit physiological stress responses before and during the delivery of bad news and experience ongoing anxiety, sometimes for a matter of days, when such interactions are perceived as not having gone well" Nelson et al 2017

disregard for parents judgement regarding the care of their child, and poor communication of important information. Such an event haunted them and complicated their grief even years later" Contro N et al. 2002

"Bereaved parents perceptions of inadequate healthcare [somatic and psychological needs, healthcare staff not fulfilling parents needs] were associated with subsequent guilt" Surkan et al 2006

"Parents perceived provider actions as insensitive and unsupportive when they were abrupt, when they were perceived as being cold and insensitive to the parent's situation, and when they laughed and joked outside the dying child's room." Brooten et al 2017

"The characteristics of physicians that were deemed most harmful to satisfying communication included having a disrespectful or arrogant attitude, not establishing a relationship with the family, breaking bad news in an insensitive manner, withholding information from parents and losing their trust, and changing a treatment course without preparing the patient and family" Hsaio et al 2007

"The meeting did not go as we hoped it would. As soon as formal introductions were exchanged, the surgeon plunged into a discussion of the

When we approached David's favorite physician to ask him to talk with our son, his body language telegraphed his discomfort. It was an uncomfortable moment for us, too. We felt that David needed professional help in dealing with his emotions, and we weren't qualified, ourselves, to handle this difficulty. But the physician didn't offer an explanation for his reluctance. We wondered how this highly skilled expert—one who could navigate through all of the medical complexities of treating cancer— could be so ill equipped in the face of a teenager's psychological struggle. With a sense of desperation, we pleaded with the physician until he relented. Three hours later, mentally exhausted, the physician "He emerged from David's room and told us that our son had many questions about death and his own mortality. The physician didn't divulge the details, but it was obvious that the discussion was much needed. We hesitantly entered David's room, not knowing how he'd reacted to the emotionally sensitive conversation. We were astonished to find him smiling and calm, in a peaceful state of mind. He was ready to play a game!" O'Shea 2013

Emotional investment in the relationship

"Provider actions perceived as caring and compassionate included nurses and doctors who cared for the child almost as if he or she was their own and when the child was nearing death called the parents to be with the child and have the opportunity to hold the child before the passing. At the time of the

complications and mortality rates associated with the risky bone marrow transplant. I turned toward David, who had covered his ears with his hands; he was looking down and shaking his head in disbelief. Joe and I were aghast. How could such a highly skilled physician be so insensitive to the power of her words and the effect they might have on our young son?" O'Shea 2013 "Parents believed that some providers had avoided talking about death or relied on a set method or technique for having an end-of-life discussion. Both avoidance and rote methods for talking about death were troubling. Parents suggested the importance of tailoring end-of-life discussions according to the needs of those participating." Robert et al 2012 "Parents were not always treated with respect and felt that staff avoided or even abandoned them (Widger & Picot, 2008; James & Johnson, 1997), leading to feelings of isolation during the palliative phase of their child's illness and that they had been physically and emotionally distance from staff with whom they had a frequent and personal relationship in the past (James & Johnson, 1997)." Melin-Johansson et al. 2014 "Suffering may result in part from a lack of recognition of the problem by the medical team. This hypothesis is supported by the finding that parents who reported that the physician was not actively involved in care at the end of life were more likely to report that their	death and immediately afterward, staff that cried and/or prayed with the parents, went to the child's funeral, and telephoned the parents after the death were perceived as caring, sensitive, and compassionate." Brooten et al 2013	

	child suffered from a great deal of pain." Wolfe et al 2000		
Care at the end of life and "palliative care"	**Expert family in a fragile, disempowered situation** "Losing a child to cancer can metaphorically be explained as being covered in a wet and dark blanket. The blanket was already present and covered the parents when the child was in its palliative phase. To see the child suffer was emotionally arduous, but facing the child's imminent death could be even more difficult. Despite the blanket, it seemed important to the participants to find the strength to talk to the child about the child's forthcoming death" Bjork et al 2016. "I always knew Daisy would die before reaching adulthood … every decision about her care was made on the basis of improving her quality of life, which meant helping her do the things she loved, such as being at home with her family, going to school and playing with friends" Nimmo 2018 "Parents felt that physicians provided a thorough assessment of the status of their child. Of interest, however, is that 20% felt that they had not been included in decisions about their child's care at EOL" Gilmer et al 2013	**Advocacy and trust** "Many parents psychologically cannot relinquish, or hand over their feeling of responsibility for the child's wellbeing, Parental feelings of guilt may, in part, be produced between the parental role and their perception of themselves as unable to act as effective caregivers under these circumstances". Surkan et al 2006 "The majority of parents remained hopeful. Hope for a cure was not associated with more long-term traumatic grief or symptoms of depression" Van der Geest 2015. "Most parents reported that health care professionals were willing to listen to their concerns (4.7 ~ 0.7) and most parents agreed that there was one specific health care professional who organized care and helped with practical issues" Van der Geest et al 2014 **Affirmation in decision making (or not)** "Perhaps more surprising is the finding that an affirmation of the rightness of decision by the doctor was highly meaningful to parents and valued by them.	**Emphasis of care placed on lessening suffering** "parents may not have to fully acknowledge their child's poor prognosis to be willing to emphasize lessening of suffering" Wolfe et al 2000 "earlier recognition of prognosis by both physicians and parents is associated with a stronger emphasis on treatment directed at lessening suffering and greater integration of palliative care" "One explanation for these findings is that communication about prognosis between physicians and parents at the time of diagnosis may be clearer than when a child's cancer is more advanced" Wolfe et al 2000 **Integration of specialist paediatric palliative care services** "Parents from the cancer group reported differing unmet needs about issues related with coping with changes in their children's ability/activity levels, knowing how to help their children cope with disease-related changes and knowing how to maintain a 'normal' lifestyle. These parents also reported the need for more access to information about palliative care, access to health professionals out of hours and

"Although the progress of the illness—the months of anxiety, hospital admissions, treatments, improvements, relapses—does, to a certain extent, prepare you for such news, it is difficult to describe the effect of it. I think crushing, stunning defeat after a prolonged, painful struggle sums it up. And of course it is the end of all hopes for recovery, when treatment stops and palliative care takes over" Darnhill S et al 2006

"After the survey, parents were also asked whether they were told their child was dying. Sixty-seven percent said they were told, and the 4 mothers who were not told indicated the child died very suddenly and without warning." Gilmer et al 2013

"Take time to explain in detail the role of each member on the health care team, including whom family members should approach for what, and what to do if there are problems. Given the central role of the primary doctor in Mexico, it is often important to explain the trade-off between having the stewardship of one primary physician versus the expertise of multiple specialists. When families report discrimination, encourage discussion. Do not dismiss their concerns with statements such as, ''That's just how Dr. Wallace acts.'' Such comments exacerbate the family's sense of disenfranchisement. Recognize that many families are reluctant to ask for help. Be proactive about offering interpreter services, written materials, and other resources in Spanish. Assessing literacy in Spanish as well English is critical to

Parents' accounts suggest that the value may derive from the fact that the doctor had witnessed the parents in their decision-making, and appreciated the magnitude of their task." Sullivan et al 2014

"Most parents agreed that health care professionals took seriously their opinion on how care for their child should be organized (4.7 ~ 0.8). Parents highly rated involvement in decision making regarding the supportive care and treatment of their child in general (4.4 ~ 1.0). Particularly during the consultation in which parents were informed that there were no more curative options, parents highly rated the attention the pediatric oncologist gave to their concerns about care for their child (4.8 ~ 0.7) and the possibility to make shared decisions (4.5 ~ 1.1). Van der Geest et al 2014

"Most paediatric palliative care decisions, however, are made jointly between mothers, fathers and physicians as opposed to a single automous decision maker" Hill et al 2013

"In relation to the more contentious issue of giving, or appearing to give, a recommendation, parents' experiences were more varied. Some parents reported that doctors gave them a recommendation: these were parents who took a guided decision-making role. Others reported that they were not given any recommendation. A small number of parents

availability of financial assistance." Monterosso et al 2007

"The results of the review highlight the importance of health-care providers understanding cultural influences specific to Chinese American parents of children with life-limiting illness. It is clear from the results that there are specific values in the Chinese culture of mainland China, Hong Kong, and Taiwan that stand in stark contrast to those of Western culture in North America. These cultural values shape Chinese American parents' experiences of their children's illnesses by mediating their perceptions, reactions, and coping behaviours. Moreover, they are reflected in clear patterns of family structure and decision making, communication and social resourcing, caregiving strain, caregiving strategies and emphases, and meaning making." Wang et al 2013.

"parents appreciated it when they felt actively involved in making decisions concerning their child's treatment and care. The parent's descriptions of an effective caregiver were highly consistent. Honesty, clinical accuracy, compassion and availability were among the most desirable caregiver traits. Contro et al. 2002

We also found that earlier discussion of hospice care was associated with a greater likelihood that parents

understanding the communication needs of the family." Contro N 2010

"Multivariate results suggest that parents with less than a high school education had decisional conflict scores (DCS) that were 13 points higher (p<0.05) than parents with some college education. In addition, parents who indicated that they had recently made a decision for their children had DCS scores that were 7 points higher (p<0.05) than parents who indicated they had not … paediatric palliative care programmes should treat parents with lower educational levels as being particularly vulnerable and should consider allocating additional resources to them when a decision for their children is imminent" Knapp et al 2010

Established relationships with healthcare professionals

"While technical care was important, a positive relationship with the primary caregivers was also considered paramount." Contro N et al 2002

Participants related that as the child neared death, they were less inclined to accept a newly initiated service or relate to a new provider. Intimacy was highly valued at the child's end of life. Trusted others were increasingly relied upon, and parents limited their child's interactions to persons well known to the

---

indicated the doctors had not simply made a recommendation, but actually made the decision. These parents felt excluded from their rightful role in decision-making. One parent said: "Well I think we were basically told that that was the best thing to do [withdraw life-sustaining treatment] …because if you just keep going then you are just being cruel to her basically. Therefore you feel like you don't have a choice". (Parent 22)" In this study, when doctors made the decision for parents, the parents viewed the doctor's action badly. It added to their difficulties after their child's death. Perhaps this came from the parent's sense of failing in their parental role, or from not being convinced of the reasons to forego treatment. Sullivan et al 2014

Shared emotional impact

"only three were directly connected to the experience of the clinician and the management of the interaction. These were 'supported', 'included' and 'trusting'. While we cannot tell what the clinicians' communication styles or processes actually were, the parents' explanations did touch on certain specific experiences. These were; being allowed to ask questions and voice concerns, being included in treatment decisions and forward planning, and having repeated opportunities to do this." Nelson et al 2017

---

would describe their child as calm and peaceful during the last month of life" Wolfe et al 2000

	family. Parents valued trusting relationships with providers. Care was considered optimal when the provider and patient had grown to know one another" Robert et al 2012	"A mother shared, "I wanted to know honestly…are you telling me we have a chance? If we don't have a chance, then tell me that we don't have a chance. Don't sugarcoat it or tell me, 'This is the best thing you should do.' Tell me everything and let me decide what the best thing is to do." Another mother wanted "more honesty from the doctors. 'Cause it seemed like they knew things, and they didn't want to say the whole truth or made it seem a little better than it was." A father stated that he was pleased with the information that was communicated to him: "Her doctors didn't really sugarcoat anything to make us believe that there was still hope when there wasn't. Through some of the therapy, I was hoping that maybe this will work…there were things I would hope for, but they (medical staff) didn't lead me on. They never, never did that…I think I was pretty informed." Steele et al 2013	

Appendix 5: Participant Information Sheets (Children and Families)

1. Story version

Do you like stories?
This is my story

Once upon a time, there was a doctor called Sarah (that's me).

She worked in a big city and cared for lots of children, and their mummies and daddies. She sometimes thought that the care they had should be better.

"How can I make this better?" thought Sarah. She thought for a long time, and thought very hard.

"I need to ask them what they think" she decided. "I will ask them to tell me their stories".

Asking people what they think or to tell their own stories like this is called a research project.

My research project is called "The Journey through Care".

The Journey through Care
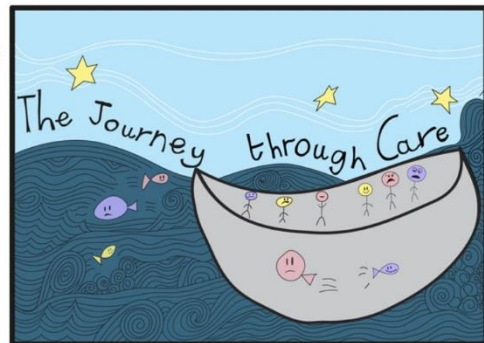
What is your story?

My project involves collecting stories. I will be asking children to tell me their stories about their care. I will ask their Mums and Dads too. The stories will be used to help to improve care for children.

If you would like to tell me your story, please ask your Mum or Dad to let me know. They can do this by filling in the "About Me" form on next page of this leaflet, and giving to your nurse or doctor, or sending it back to me. I will be in touch with you soon.

Thank-you!

My details:

Sarah Mitchell
University of Warwick Medical School
Gibbet Hill Road Coventry CV4 7AL

"About Me" form
Please could you answer the questions below about you and your family?
Please return the form to your nurse or doctor, or directly to me in the envelope provided. Or email me with your details █████████████████████, or phone or text ███████████ You can contact me by email, phone or text and I will get back to you as soon as possible.

My diagnosis:

My Parents:

My Address:

How to get in touch with us: (Preferred contact details)
Tel:

Mobile tel:

Email:

When it is best to get in touch with us (please tick):

	Morning	Afternoon	Evening
Monday			
Tuesday			
Wednesday			
Thursday			
Friday			
Saturday			
Sunday			

Thank-you

Sarah is funded by a National Institute for Health Research Doctoral Research Fellowship .

This leaflet has been designed with the help of young people's groups from Acorns Children's Hospice and Birmingham Children's Hospital.

WARWICK
THE UNIVERSITY OF WARWICK

Birmingham Community Healthcare **NHS**
NHS Trust

Birmingham Children's **NHS**
Hospital
NHS Foundation Trust

2. Information Sheet for Children and Young People
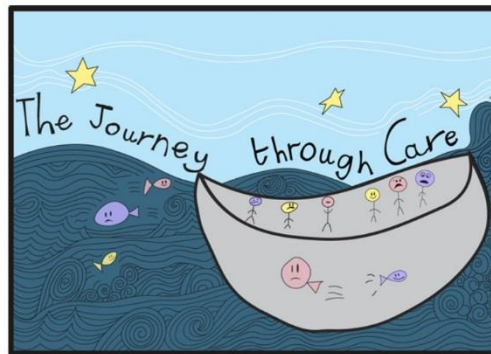
Project Information Sheet

Hello

My name is Sarah Mitchell. I am a student at the University of Warwick, and I also work as a GP. I would like to invite you to take part in my research project.

This leaflet will provide you with more information about the project.

What is the project about?

The project is about understanding what children, young people and families think about the care they receive from the NHS when they are seriously unwell or have a condition that may not get better, and to consider how these services can be improved.

How many interviews?

It might take some time to tell your story. You might think of new things you want to say. We can have 1, 2 or 3 interviews. It's up to you.

Do I have to

No, it is up to want to take pa change your mi without giving a re

What will happen if I decide to take part?

- First we will arrange a time to talk about the study in more detail.
- There is a form to sign if you decide to take part.
- Your parents will also be asked to sign a form.
- After that, we will arrange an interview, with or without your parents.
- In the interview, I will ask you to tell me your own story about being unwell, about what is important to you and about the people who help or care for you.
- You can say whatever you want, and as much as you want.

Why have I been asked to take part?

You have important views on what it is like to receive health services and what would make them better.

take part?

...you whether you
...rt, and you can
...nd at any time
...eason.

What will happen to the information?

The interviews will be recorded. The information will be analysed by researchers and used to show how services can be improved. Nobody else will be able to know what you have said, except:

- If there are things that are very difficult to discuss and we find you need some extra support. If this is the case we will talk together and with your parents about what you need and how best to organise this.

- If there are any concerns about your safety.

Why should I bother taking part?

Imagine if you could change healthcare services so that they work better, for you and your family and for many other children and young people who will need them in the future. That would be amazing.

It's hard to make change happen. Research can help. By taking part in this study you will be helping to work towards that important change.

How can I take part?

Please complete the "About Me" form and send it back to me in the envelope provided OR hand it back to your nurse, and I will be in touch with you soon to provide any more details that you need and discuss whether you would be willing to participate in the study:

Thank-you!

"About Me" form

Please could you answer the questions below about you and your family?

Please return the form to your nurse or doctor, or directly to me in the envelope provided. Or email me with your details █████████████████████████, or phone or text ████████████ and I will get back to you as soon as possible. Thank-you

My Name:

My diagnosis:

My Parents:

My Address:

How to get in touch with us: (Preferred contact details)

Tel:

Mobile tel:

Email:

When it is best to get in touch with us (please tick):

	Morning	Afternoon	Evening
Monday			
Tuesday			
Wednesday			
Thursday			
Friday			
Saturday			
Sunday			

The Journey through Care:
Study Information

This information sheet is about a research project that is being completed by Dr Sarah Mitchell at the University of Warwick. The project has been designed to investigate how the NHS provides care to children and young people who are living with serious conditions which may or may not get better; conditions that could be considered life-limiting or life-threatening. The aim of the project is to consider how healthcare services can be improved in the future.

Please read this information sheet to help you decide whether you and / or your child would like to take part. The information sheet will tell you what taking part would mean for you.

What is the project about?
The aim of the project is to understand what children, young people and their families think about the healthcare services that they receive. The project will involve interviews with children and young people and family members who live in the same household. The aim is to consider what works, what doesn't work, and how NHS services can meet the growing needs of seriously ill children, young people and their families in the future.

Why have I been asked to take part?
Children and young people and their families are increasingly sharing their personal stories about illness and their experiences of healthcare services. One example of this are the WellChild Family Bloggers (https://www.wellchild.org.uk/families-area/connect-with-others/). Maybe you have a story too. This research project recognizes that your ideas and experiences, and those of your child, are important. The project is particularly about whether NHS services work, or not, and which services are the most valuable to you.

What is the aim of the project?
The project will collect information (stories) about how services are working now. The stories will be carefully analysed and used to help to improve services for children and young people and their families in the future. A new model of care will be proposed using the research findings which can be used by managers and policy makers. The research will also be written up as academic papers and will be presented at conferences, and local, regional and national service and policy development meetings.

What will happen if my child or I decide to take part?

If you could tell your story and would like to take part in the project, please send the attached contact details form back to me. I will then contact you and arrange a time for an interview. I will answer any other questions you have about the study, ask you to sign a consent or agreement form, and arrange an interview with you and your child if that's possible. If you can support your child to take part, there will be a separate form to sign.

Interviews will be arranged at a place and time that is convenient for you. It is anticipated that an interview will last anywhere from 20 – 40 minutes, but there will be no time limit.

You can take part in one, two or three interviews over time if you would like to. This will allow you to gradually tell your story in more detail.

What will happen to the information that is collected?

The interview will, with your consent, be recorded, using a small digital audio recorder. The recordings will be typed up and used later for data analysis by the research team. Your identity will be kept confidential. Any personal information provided or information that identifies other people will be anonymised. Anonymised quotes may be used in publications, for future research and in the development of future educational resources.

The information you give will be only be seen by the researchers, unless either of the following exceptional circumstances arise:

1. If there are things that are very difficult to discuss and you that you might need extra support to deal with. If this is the case we will agree how best to organise this.
2. Although not anticipated, should any examples of professional misconduct, negligence or child protection concern be disclosed, the researcher is bound by the professional code of conduct to report this information to the Clinical Director of the Trust.

Can I have a copy of the recording?

You are welcome to have a copy of the typed out version of the recording of your interviews.

Do I / we have to take part?

No. Participation in this research is voluntary. If you agree to take part, you can change your mind at any time and you do not have to give a reason. If you wish to withdraw consent following the interview, all data relating to the interview, including recordings and transcripts will be destroyed and not included in the study. Your decision about whether to take part or not will in no way influence the care that you receive.

Who can I contact for more details?
If you would like to know more about this project, or if you have any questions please contact:
Sarah Mitchell (Clinical Research Fellow)
University of Warwick Medical School, Gibbet Hill Road, Coventry, CV4 7AL
Email: ▮▮▮▮▮▮▮▮▮▮▮▮▮▮▮▮▮▮ Tel: ▮▮▮▮▮▮▮▮
You can contact me by email, phone or text and I will get back to you as soon as possible.

What if there is a problem?
If a problem arises or you wish to make a complaint about the conduct of this study or any people involved in it, you may do so by writing to or speaking to the researcher, Sarah Mitchell, who will do her best to answer your queries. If you would prefer not to raise your concern with Sarah, please contact Prof Jeremy Dale (email: ▮▮▮▮▮▮▮▮▮▮▮▮▮▮▮▮, tel: ▮▮▮▮▮▮▮).

Thank-you to the young people's and parents groups from Acorns Children's Hospice, Birmingham Children's Hospital and A Child of Mine who helped to design this leaflet.

324

Appendix 6: Participant Information Sheets (Focus Groups)
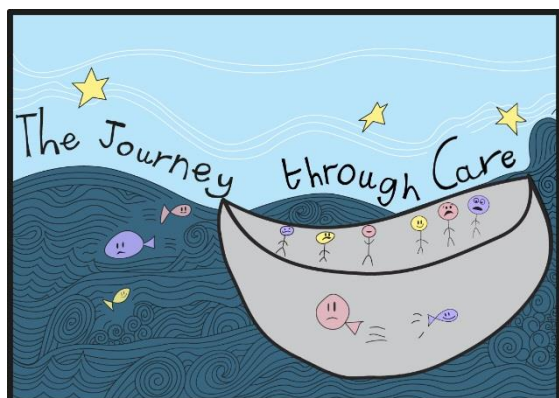
What is Palliative Care for Children and how should it be delivered?

Information Sheet for Networks

This information sheet is about a research project which is being carried out by Dr Sarah Mitchell at the University of Warwick. The project is about "palliative care" services, and to consider how these might be improved in the future. The project is funded by a National Institute for Health Research (NIHR) Doctoral Research Fellowship.

You are invited to take part in a group discussion about the initial finding. Please read this information sheet to help you decide whether you would like to take part.

What is project about?

The aim of the project is to understand what children, young people and their parents think about the healthcare services that they receive, and "palliative care" in particular. Children and young people, parents, staff from health services, and commissioners will all be interviewed about this. The aim is to consider what works, what doesn't work, and how NHS services can meet the growing needs of children, young people and families in the future.

Why have I been asked to take part?

As a member of a palliative care network you will have important ideas and experiences of services in your area, including things that work and which services are the most valuable. You may also have ideas about how this might be improved, for children and young people, families and staff.

The project is collecting important information about how services are working now (the research findings). The initial findings of the project will be presented to you today. As an expert in your region, your opinion is valuable in terms of trying to establish whether the initial findings are similar to your experience of services, or not.

It is hoped that the findings of the study will be used to help to improve services for children and young people and their families in the future, through presentation as papers and at conferences, and at relevant local, regional and national service and policy development meetings.

What will happen if I decide to take part?

I would like to audio-record the group discussion about the study findings, in order to keep a record of those discussions. The recordings will be transcribed and used as part of the data analysis process. If you agree to take part, please sign a consent form.

Your identity will be kept confidential. Any personal information provided or information that identifies other people will be anonymised. Anonymised quotes may be used in publications, for future research and in the development of future educational resources.

Do I have to take part?

No. Participation is voluntary. If you agree to take part, you can change your mind at any time and you do not have to give a reason. If you wish to withdraw consent, any contribution that you have made to the discussions will be removed from the transcripts and not included in the study.

Who can I contact for more details?

If you would like to know more about this project, or if you have any questions please contact:

Sarah Mitchell (Clinical Research Fellow)

University of Warwick Medical School, Gibbet Hill Road, Coventry, CV4 7AL

Email: ██████████████████ Tel:

What if there is a problem?

If a problem arises or you wish to make a complaint about the conduct of this study or any people involved in it, you may do so by writing to or speaking to the researcher, Sarah Mitchell, who will do her best to answer your queries. If you would prefer not to raise your concern with Sarah, please contact Prof Jeremy Dale (email: ██████████████████, tel: █████████).

Appendix 7: Dissemination and impact summary

The PhD dissemination activity has included the following awards and invited talks:

2019	Invited international keynote speaker: Bioethics conference, Royal Children's Hospital, Melbourne (September 2019)
2018	Example of impact: Systematic review referenced in the 4th edition of Together for Short Lives Guide to Children's Palliative Care.
2018	RCPCH Conference workshop with PPI group members
2017	NIHR CRN West Midlands Awards: Highly Commended for Involving Patients & Users
2017	Oral presentations with PPI group members: International Children's Palliative Care Network and Society of Academic Primary Care Conferences.
2017	Poster Prize: 8th International Children's Palliative Care Network Conference
2017	Speaker on the NHS England national webinar for Paediatric Palliative Care
2017	Panel discussion member (invited): "Untangling the knotty problems". Together for Short Lives National Conference.
2016	Chair (invited), RCGP / Marie Curie Primary Palliative Care Research Development Day
2015	Travel Bursary: 6th International Children's Palliative Care Network Conference
2015	Shortlisted for Health Services Journal Rising Stars Award
2015	Plenary (invited): "From research to policy to practice and back again: A marathon not a sprint", Together for Short Lives 2015 National Conference.

I am committed to ensuring that research informs policy and practice, and became Clinical Lead for the Birmingham Cross City and South Central Clinical Commissioning Groups (CCGs) from 2014-2017. I led on the development of an evidence-based Palliative and End of Life Care Strategy for the city, which proposed integrated palliative care through commissioning and innovative contracting across primary, secondary and community care and voluntary sector organisations. I also led on the development of a strategy for Children's Palliative and End of Life Care. Birmingham and Solihull is now a national exemplar, with palliative care for children named as a Sustainability and Transformation Partnership (STP) area priority.

A family move to Sheffield in the summer of 2018 has brought more opportunities. I have become the Macmillan GP facilitator for the city, providing support to GP practices to achieve their quality improvement targets for the Quality and Outcomes framework 2019/20, and working closely with local commissioners. I have also become the Clinical Lead for Children and Young People. Priority for the commissioners for children's services in Sheffield include improving the community service offer for all children, but particularly those with complex and palliative care needs. I am the GP representative on the Association of Paediatric Palliative Medicine Executive Committee, a role in which I will work to support GPs and doctors working in children's hospices, and GPs who may be involved in the delivery of palliative care to children in their practice. I am also a member of the Executive Committee of the Yorkshire and Humber Children's Palliative Care Network, and the NHS England Paediatric Medicine National Clinical Reference Group, both roles through which I can provide advice and insight into work to improve the integration of palliative care in the care of children with complex and palliative care needs.

Appendix 8: PhD comic strip presentation

The slides below have been used to disseminate the findings of the research and local, regional, national and international conferences. They were created with the advice of the PPI group, and using an online application, www.storyboardthat.com.

Slide 1: Aims of the research – to capture the views of children, and take a realist approach
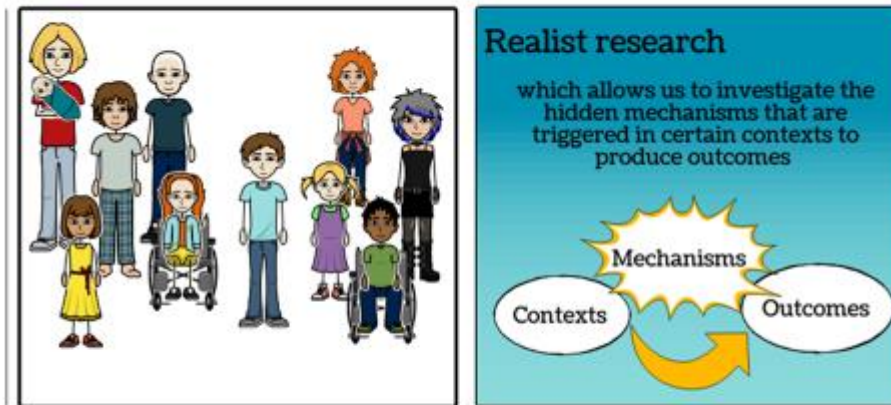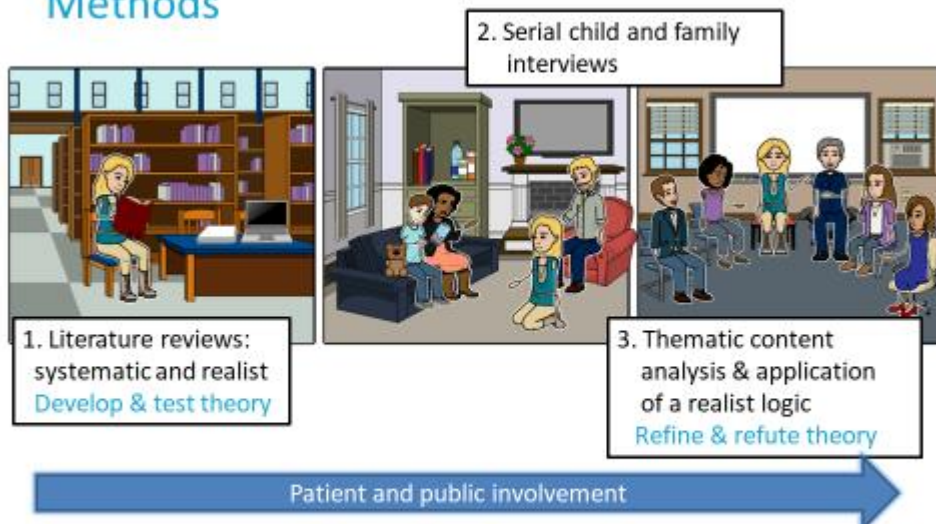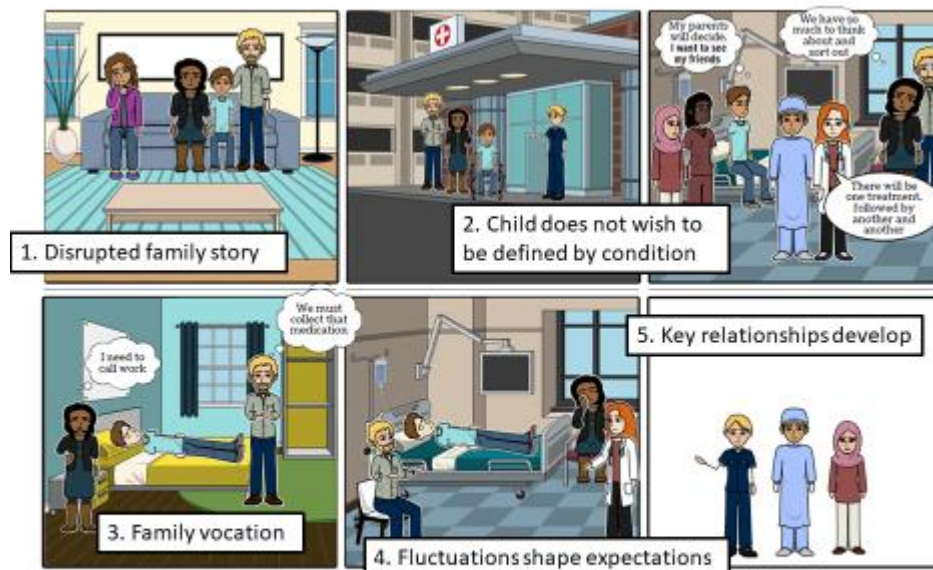

Slide 2: Methods included literature reviews, serial child and family interviews, and focus groups with healthcare professionals

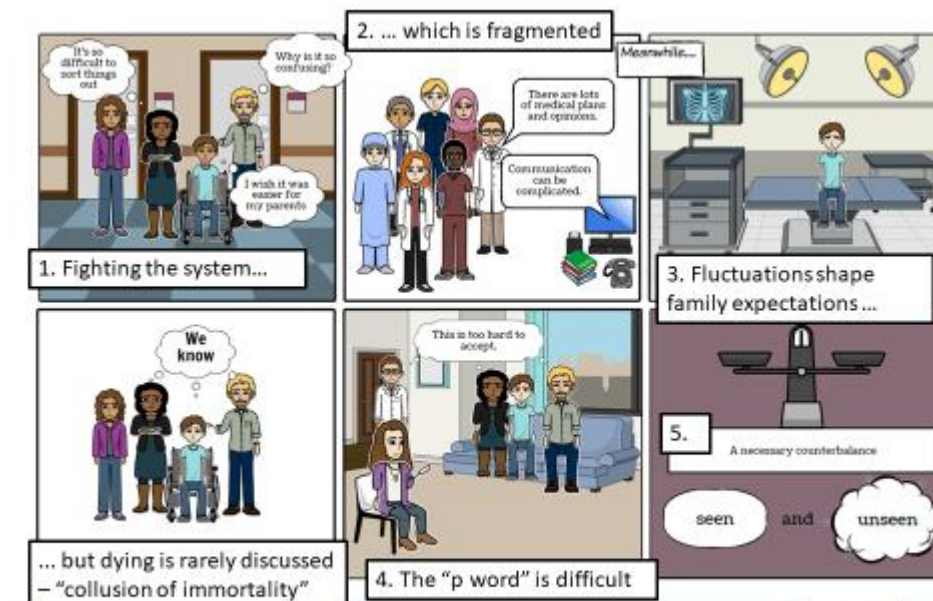Thematic analysis 1: The micro-system

https://www.storyboardthat.com/

Findings 2: Meso and macro systems

https://www.storyboardthat.com/

Programme theory

Level	Micro	Meso	Macro
Context: family situation	Expert family Vulnerable situation Child with own views	Increasing complexity Significant uncertainty Fragmented system	Complex healthcare system
Context: resource	Connection Motivated professionals	Specialist paediatric palliative care team	Accountable senior leaders
Hidden mechs	Advocacy Development of trust Listening to listen Ability to bear witness to child and family situation	Role-modelling Legitimising approach Challenging the collusion of immortality	Commitment Shared vision Role-modelling
Outcome	Trusted relationships Family feel respected, heard, "looked after" Child's priorities identified and heard	Change in behaviours of individuals Early identification of palliative care need	Service development Organisational culture change

"Being alongside"
Role-modelling
Legitimising approach

https://www.storyboardthat.com/

Slide 6: Conclusion and policy-relevant recommendations

Turning policy into reality

Across the healthcare system:

► Prioritise palliative care for children and families

► Children are highly individual and do not always fit criteria

► Prioritise relationships with healthcare professionals (and the contexts that enable these)

► Nurture and support professionals

► Develop palliative care within other specialities

► Organisational culture has an impact

► Committed system leaders are required

https://www.storyboardthat.com/