



# A mixed methods study exploring how parents of children with cancer manage their child's cancer pain at home

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# Abstract

**Background:** Children with cancer experience pain throughout their cancer trajectory, which has short- and long-term negative consequences, both physically and psychologically. Treatment advances have increased ambulatory care, enabling children to spend more time at home. This leads to a shift in pain management responsibility from healthcare professionals who have experience in pain management, to parents, most of whom do not. Little is known about parents' pain management abilities at home.

**Aim:** To understand how parents of children with cancer manage their child's cancer pain at home.

**Methods:** Guided by the Theory of Planned Behaviour, a convergent, parallel mixed methods design was used for the primary purpose of complementarity. Participants were parents of children with cancer on active treatment, recruited from one tertiary cancer centre in the South of England. Convenience sampling was used to recruit participants to complete surveys and pain diaries. Purposive sampling was used for interviews. Surveys measured parents' attitudes toward pain medications and their misconceptions regarding pain assessment. Pain diaries gathered baseline data on pain intensity, cause, location, and parent interventions at home. Interviews enabled deeper understanding of children's pain manifestation, as well as parents' pain assessment and interventions. Surveys and pain diaries were analysed using descriptive and inferential statistics. Thematic analysis was used to analyse interview data. Each dataset was analysed separately and then integrated.

**Results:** Integration of the datasets revealed that most of the time, children were not in pain at home. However, most children experienced episodes of clinically significant pain. Parents combine different types of information to effectively assess their child's pain at home. Parents frequently under-medicated their child's pain but used a variety of non-pharmacological interventions to manage their child's pain at home. Paracetamol, ibuprofen, codeine, and morphine administration all have undesirable consequences for children with cancer who frequently found pharmacological interventions to be unpalatable. This left parents with an "empty toolbox" of pain management interventions which they restocked with non-pharmacological interventions.

**Contribution to knowledge:** Using mixed methods, this research took a holistic approach to investigating parents' management of children's cancer pain at home. This research suggests children with cancer have heterogeneous pain trajectories but due to the unpredictability of pain at home, it is important all parents of children with cancer are prepared for their pain management role. Parents' use different types of information to assess pain. This constitutes a bundled approach to pain assessment. This approach may be helpful in clinical and research contexts. This research is the first to outline practical barriers to pharmacological interventions and the subsequent importance of non-pharmacological interventions for parents managing their child's cancer pain at home.

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## **List of Abbreviations**

ATC – Around the Clock

CBT – Cognitive Behavioural Therapy

CRD – The Centre for Reviews and Dissemination

HCP – Healthcare professional

NHS – National Health Service

NICE – National Institute for Health and Care Excellence

NRS – Numerical Rating Scale

PBC – Perceived Behavioural Control

PPI – Patient and Public Involvement

QOL – Quality of Life

RCT – Randomised Controlled Trial

SCT – Social Cognitive Theory

TPB – Theory of Planned Behaviour

TRA – Theory of Reasoned Action

UK – United Kingdom

US – United States

WBFPS – Wong-Baker Faces Pain Scale

WHO – World Health Organisation

# Chapter 1. Introduction

In 2011, a publication by Fortier and colleagues called for researchers to investigate children's cancer pain in the home setting (Fortier *et al.*, 2011b). They presented the argument that whilst there is evidence children with cancer experience pain at home, and that parallel bodies of literature suggest parents struggle with managing pain, insufficient evidence exists to design interventions. Research presented in this thesis focuses on bringing knowledge to the point where there is sufficient evidence for researchers to begin designing interventions to reduce the amount of pain experienced by children with cancer at home.

## 1.1 Overview of introduction chapter

To support the development of this knowledge, this chapter sets out the current body of understanding. It opens with an outline of the clinical background to demonstrate the increasing role parents take in managing pain. Trends in the location and form of treatment for children with cancer are described. This is followed by symptoms of cancer, cancer pain prevalence, and consequences of pain which establishes the relationship between cancer and pain. Pain management is discussed next including the analgesic context of children's cancer pain management and parents' management of children's pain. This clinical background leads to a presentation of the rationale for the study and the imperative for research in this area provided by government and national policy. Concluding sections provide a personal perspective, clarification of terminology, and an overview of thesis organisation to orient readers to the remainder of the thesis.

## 1.2 Clinical background

### 1.2.1 Cancer trends

In the United Kingdom (UK), recent figures suggest 1,821 children (Cancer Research UK, 2015a) and 2,630 teenagers and young adults (Cancer Research UK, 2015b) develop cancer annually. Since 1975, death rates from childhood cancer have declined by 50% (Ward *et al.*, 2014) and survival rates in England and Wales have increased to 82% for children and 84% for teenagers and young adults; figures are similar in the United States (US) (National Cancer Institute, 2015). These growing survival rates are in part, a result of increasingly aggressive systemic anti-cancer therapies (Clarke *et al.*, 2005; Fortier *et al.*, 2014).

In line with patient preferences (Jibb *et al.*, 2018), cancer care is increasingly delivered in outpatient clinics which leads to children with cancer spending more time at home with their parents, and less time in hospital (Fortier *et al.*, 2011b; Kazak and Noll, 2015). This trend towards outpatient treatment and restriction of hospitalisations to complications only is growing internationally (Kars *et al.*, 2008; Kazak and Noll, 2015). Government policy in England is clear that children with cancer should be admitted to hospital only as required, and discharge should be facilitated as soon as possible (NHS England, 2013). Patients and families show preference for this change in treatment location which is associated with improved quality of life (QOL) (Fortier *et al.*, 2014; Jibb *et al.*, 2018). Many authors recognise a subsequent shift in responsibility for symptom management from healthcare professionals (HCP), who are trained in pain management, to parents most of whom are not trained (Fortier *et al.*, 2014; Kazak and Noll, 2015; Twycross *et al.*, 2015b). This shift incurs a heavy burden on parents who are coping with this in addition to multiple new tasks which come with caring for a child with cancer (Clarke *et al.*, 2005; Flury *et al.*, 2011; Molinaro and Fletcher, 2018).

### **1.3 Symptoms of cancer**

Advances in symptom management have not matched progress in treatments resulting in children experiencing severe side-effects which reduce QOL (Dupuis *et al.*, 2016; Sato *et al.*, 2014; Tutelman *et al.*, 2018). Side-effects are wide ranging and include reductions in physical wellbeing, psychological wellbeing, and autonomy (Bettle, 2015; Flury *et al.*, 2011; Williams and McCarthy, 2015). This research aims to contribute knowledge which would support development of interventions to address this imbalance between treatment and symptom management in the specific case of pain-management at home.

#### **1.3.1 Pain prevalence and variables**

Literature frequently cites pain as the most commonly reported, bothersome, and distressing side-effect of children's cancer treatment (Hedén *et al.*, 2013; McClain and Suresh, 2011; Olson and Amari, 2015; Van Cleve *et al.*, 2012). Pain continues throughout the cancer trajectory (Fortier *et al.*, 2014; Hedén *et al.*, 2013; Olson and Amari, 2015; Van Cleve *et al.*, 2012), and is associated with a reduced QOL (Van Cleve *et al.*, 2012). In the palliative phase of cancer, complications which prevent effective pain management are present (Marks *et al.*, 2013), and pain management focuses on improving QOL rather than cure which is the opposite to previous phases of the disease (Roza *et al.*, 2014). For these reasons, this thesis describes research which does not focus on pain in the palliative phases but focuses on pain management at home whilst children are on active treatment.

Numerous studies have attempted to correlate pain in children with cancer with other variables including age, diagnosis, and gender. Studies into the effect of age on children's cancer pain experience have mixed results (Twycross *et al.*, 2015b). One study found symptom burden, as reported by parents, did not differ with age (Hedén *et al.*, 2013). Girls with cancer have reported higher pain intensity, although this retrospective finding may be reflective of gender differences in pain recall (Hechler *et al.*, 2009). Other studies found no gender differences in pain experienced by children with cancer (Hedén *et al.*, 2013; Van Cleve *et al.*, 2012; Walco *et al.*, 2005). Symptoms experienced by children with cancer do not differ with diagnosis (Hedén *et al.*, 2013). Although children with brain tumours may be at increased risk of headaches (Klitbo *et al.*, 2011; Sato *et al.*, 2014), few studies provide sufficient diagnosis information to allow such conclusions (Olson and Amari, 2015). Children's pain was highest (69%) one week into a cycle of chemotherapy compared to prior to (62%) and two weeks after chemotherapy (52%) which suggests pain may differ depending on stage of treatment (Baggott *et al.*, 2010). There are difficulties in providing a single figure of cancer pain prevalence in children due to differing study populations, timeframes and aspects of pain (Twycross *et al.*, 2015b).

### ***1.3.2 Causes of pain in cancer***

Children with cancer experience pain originating from three sources: procedures; the disease itself; and side-effects of treatment (Fortier *et al.*, 2014; Hedström *et al.*, 2003; Olson and Amari, 2015; Twycross *et al.*, 2015b). Painful procedures include surgery, lumbar punctures, venepunctures and bone marrow aspirations (Fortier *et al.*, 2014). These procedures typically take place within the hospital environment where HCPs are responsible for pain management. Research in this thesis will, therefore, focus primarily on pain from the disease itself and from side-effects of treatment, which are experienced more regularly at home.

The disease itself can cause pain directly and indirectly (Gupta, 2018). Direct mechanisms include extension of tumours into soft tissue, visceral or bone involvement, nerve compression or nerve injury and raised intracranial pressure (Oakes, 2011). Indirectly, cancer can cause pain through mechanisms such as muscle spasm, constipation, and concurrent disorders. Various cancer treatments cause pain for children: chemotherapy can result in mucositis and infections; radiation can cause burns, dermatitis, and myelopathy; and surgical interventions can result in pain during recovery (Gupta, 2018; Van Cleve *et al.*, 2004; WHO, 2012).



### **1.3.3 Consequences of pain**

Addressing children's cancer pain is important as experiences of childhood pain have lasting negative effects (Burke *et al.*, 2017; von Baeyer *et al.*, 2004). Pain produces a biological stress response (Sinatra, 2010; Tennant, 2013) which can negatively impact an individual's ability to eat, sleep, think, and interact with others (Berger *et al.*, 2013). In adults with cancer, pain reduces QOL and is associated with mental health conditions such as depression, anxiety, and posttraumatic stress (Davis and Walsh, 2004). In children with cancer, pain impairs QOL (Jibb *et al.*, 2015) and delays recovery (Shepherd *et al.*, 2010). At home, children with cancer report that pain negatively affected their sleep, as well as home, school, and social functioning (Fortier *et al.*, 2014). Recent evidence suggests cancer pain can become chronic and continue to be problematic even into survivorship (Alberts *et al.*, 2018; Tutelman *et al.*, 2018).

## **1.4 Pain management**

### **1.4.1 Analgesic context of children's cancer pain management**

For much of their cancer journey, as a side-effect of treatment, children with cancer lack necessary white blood cells to fight infection – a status known as neutropenic. During this time, children are vulnerable to infections, which are potentially fatal if not detected early and treated with intravenous antibiotics (Cheng and Tattermusch, 2014). This has implications for pain management as, due to its antipyretic effect, paracetamol risks masking a raised temperature, which may be a parents' only sign of infection at home (Oberoi *et al.*, 2013). When children are neutropenic, protocols state that parents can administer paracetamol safely for pain if their child's temperature is within normal ranges (Cheng and Tattermusch, 2014). Ibuprofen is contraindicated for children with cancer due to its association with bleeding (Cheng and Tattermusch, 2014; Hanmod and Gera, 2016). Of note, ibuprofen also holds antipyretic properties so would have similar implications to paracetamol for masking a raised temperature, but this is not its primary reason for being contraindicated.

Guidance released by the Medicines and Healthcare Products Regulatory Agency in 2013 indicates codeine is no longer recommended for children (MHRA, 2013) and many institutions have withdrawn it from their formularies (Andrzejowski and Carroll, 2016; Cheng and Tattermusch, 2014; Hanmod and Gera, 2016). Morphine is frequently recommended for children's cancer pain, but undesirable side-effects including constipation and nausea are acknowledged (Cheng and Tattermusch, 2014; Hanmod and Gera, 2016). Guidelines recommend administration of laxatives and antiemetics alongside morphine (Cheng and Tattermusch, 2014; Hanmod and Gera, 2016). A recent Cochrane review concluded morphine was an effective analgesic drug for cancer pain in

adults (Wiffen *et al.*, 2016). However due to lack of evidence, a similar review could not conclude in favour of morphine efficacy for children's cancer pain (Wiffen *et al.*, 2017).

Non-pharmacological interventions, also termed complementary and alternative medicines, or physical and psychological interventions, are recognised in oncology protocols as alternative methods of pain management for children with cancer (Cheng and Tattermusch, 2014). Evidence in support of non-pharmacological interventions for cancer pain is growing but remains weak (Bao *et al.*, 2016; Jibb *et al.*, 2015). In clinical practice, evidence suggests these interventions are under-utilised (Plummer *et al.*, 2017). The extent to which these interventions are used by parents managing children's pain at home is unknown as previous research has focused solely on pharmacological interventions (Fortier *et al.*, 2014).

#### **1.4.2 Parents' management of pain**

Literature examining pain management abilities of parents, and effectiveness of interventions, to improve pain management at home in children from non-cancer populations, highlights factors which may affect parents' management of children's cancer pain at home (Twycross *et al.*, 2015b). Twycross *et al.* suggested that the extensive literature on parents' management of children's postoperative pain may be a helpful indicator of pain management abilities of parents of children with cancer (2015b). Several studies found that while parents are able to assess pain postoperatively, this does not always result in appropriate pain management (Fortier *et al.*, 2009; Kankkunen *et al.*, 2009; Longard *et al.*, 2016). Research revealed attitudinal barriers in parents as well as a fear of side-effects and addiction potential of analgesic drugs (Twycross *et al.*, 2015d). In one study, parents (n=315) agreed postoperative pain management was their responsibility but reported finding it challenging (Kankkunen *et al.*, 2003). A recent literature review concluded that behavioural approaches may be most effective at improving parents' management of children's postoperative pain (Chorney *et al.*, 2014). Therefore, the theoretical framework for this study, described in Chapter 4, was drawn from behavioural literature.

### **1.5 Government and national policy**

Health care priorities in England focus on improving QOL, improving symptom management, and supporting carers of patients with cancer (Corner & Wagland, 2013; Department of Health, 2015; NICE, 2005). Recent government policy establishes cancer patients' QOL as one of six key action areas (Department of Health, 2015) reflective of the cultural shift in cancer care from clinical outcomes towards patient experience (Department of Health, 2011).

Policy acknowledges the challenges of ensuring adequate pain management at home (NHS Quality Improvement Scotland, 2009). Patient and carer held barriers to pain management include fear of addiction, tolerance of pain medication, fatalistic attitudes to pain, and lack of knowledge regarding analgesic drugs (British Pain Society, 2010). Government and other national bodies emphasise the importance of families in improving pain management (Corner & Wagland, 2013; Department of Health, 2011; NICE, 2014) in both adults (NHS Quality Improvement Scotland, 2009) and children with cancer (Royal College of Nursing, 2009). The Department of Health advocates for interventions to increase the ability of patients with cancer to manage their symptoms at home (Corner & Wagland, 2013; Department of Health, 2011). The National Institute for Health and Care Excellence (NICE) guidelines on Improving Outcomes in Children and Young People with Cancer call for research into the management of symptoms such as pain which will aid discharge and increase QOL (NICE, 2005). Research in this thesis was designed to address the needs outlined in these policies.

## **1.6 Personal perspective**

The author is of central importance to research, so it is important to reflect on my<sup>1</sup> own position at the start of this research. My interest in this topic has been shaped by my research and clinical backgrounds. In 2013, as a research assistant, I was part of a group of researchers who conducted a rapid review examining children's cancer pain with a focus on cancer pain at home (Twycross *et al.*, 2015b). Results of this review are described in detail in Chapter 2. In conducting this research, I was exposed to researchers with expertise in both children's pain and children's cancer. I explored the negative effects pain has on children physically and psychologically in both the short- and long-term and how postoperative literature describes parents as unprepared for effective pain management.

Through this exposure I learned that due to medical advancement: (1) Survival rates for cancer are increasing and there is a shift in focus from cure, to improving quality of life; (2) Newer, more aggressive therapies are associated with an increase in treatment side-effects including pain; (3) Children with cancer spend more time at home cared for by their parents; and (4) Modern pain management interventions are growing in efficacy and as a result more pain in children can be controlled. Postoperative pain research frequently describes parents as holding negative attitudes and misconceptions regarding pain management and struggling to manage their child's pain at

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<sup>1</sup> The personal pronoun has been used in this section as it provides my personal perspective.

home. Although I tried to be open-minded, at the start of this research, I expected to find similar negative attitudes in my doctoral studies.

Due to my clinical role, I held an element of the emic perspective on the setting of this research. I had some understanding of the disease, treatment, and side-effects. However, parents are the focus of this research and I had no parenting experience so was taking primarily an etic perspective for much of this research. My main experience of parenting came from my own upbringing in which I had been relatively healthy and in little need of either healthcare interventions or pain management interventions.

As a children's cancer nurse, I saw children and their families go through trauma of cancer diagnosis and subsequent treatment. Many negative aspects of the journey they were about to undertake could not be mitigated but due to my research experience, I knew that there were options for alleviating pain. At the commencement of my doctoral studies, I found no interventions to support parents in managing children's cancer pain at home. As I explored the literature, I discovered that it did not provide sufficient definition of this problem to enable intervention development. My doctoral studies developed from an awareness of the dissonance between what was possible in terms of pain management and what I saw from the patients and families I cared for.

## **1.7 Thesis terminology**

In this section, several aspects of terminology will be clarified for the purpose of this thesis. In this thesis, "parent" refers to mothers, fathers, guardians or any individual with primary caregiving responsibility for a child. In this thesis "pain manifestation" refers to pain intensity, prevalence, cause, and location. A "child", in this thesis, is defined as an individual aged from birth to one day before their 17<sup>th</sup> birthday. A definition of "home" in this thesis is any context outside the healthcare setting where pain is not primarily managed by healthcare professionals.

Thirdly, the interventions used by parents have been grouped under terms which vary between data collection methods. In the pain diary results, "pharmacological interventions" include analgesic drugs, topical creams, antiemetic drugs, laxatives, and antidiarrheal medications. These interventions are separated from "non-pharmacological interventions" which include all other responses to pain, for example, cuddles, distraction, and massage. These groupings are reflective of how parents recorded responses to their child's pain in pain diaries as parents provided dosing for

pharmacological interventions but not for non-pharmacological interventions. Slightly different terminology is used when describing the findings of the interview where “pharmacological interventions” refers only to analgesic drug interventions including paracetamol, ibuprofen, morphine, codeine, and adjuvant drugs including pregabalin and gabapentin. “Non-pharmacological interventions” include physical strategies such as massage, psychological strategies such as distraction, non-analgesic drugs such as ondansetron, and other interventions such as topical analgesics. Although this category includes some pharmacological interventions (i.e. non-analgesic drugs), parents’ attitudes and description of analgesic drug interventions differentiated them from all other interventions, so a pragmatic decision was made to group interventions accordingly when analysing interview data. Table 1.1 summarises how terminology has been used and displays examples of how interventions have been classified in pain diary and interview results.

Table 1.1: Terminology definition of pharmacological and non-pharmacological interventions

	<b>Pharmacological interventions</b>	<b>Non-pharmacological interventions</b>
<b>Pain diary results</b>	Analgesic drugs, topical creams, antiemetics, laxatives, and antidiarrheal drugs.	All other interventions including: cuddles, distraction, massage.
<b>Interview results</b>	Paracetamol, ibuprofen, morphine, codeine, pregabalin, and gabapentin.	All other interventions including: massage, distraction, non-analgesic drugs (e.g. ondansetron), topical analgesics.

## 1.8 Organisation of the thesis

This chapter provided an overview of the clinical and political background to the thesis. Chapter 2 provides a literature review which aimed to ascertain current understanding of children’s cancer pain at home and parents’ management of that pain. The review was an integration of a rapid review conducted in 2013 in preparation for doctoral studies and an update of that review conducted in 2016 at commencement of doctoral studies presented in a single report. This report concludes that whilst there is growing evidence that children with cancer experience pain, insufficient evidence exists to develop interventions to support parents managing their child’s pain at home.

Chapter 3 describes the findings of a published integrative literature review with a twofold aim. Firstly, it aimed to identify interventions which support parents in managing their child’s pain at home. This investigation considered pain caused by any acute or chronic condition. Secondly, it aimed to ascertain which aspects of interventions increase the likelihood of their success. The

findings of this review supports empirical work described within the thesis and enables researchers to begin development of effective interventions to support parents managing their child's cancer pain at home.

Chapter 4 documents the search for a suitable theoretical framework to guide further understanding of this research. Two theoretical frameworks are described in detail. Analysis of the suitability of these frameworks is conducted and rationale for choosing the Theory of Planned Behaviour is provided. This chapter finishes with a description of how the theoretical frameworks have been applied to this research.

Chapter 5 outlines the methodology for this convergent parallel mixed methods study which utilised surveys, pain diaries and interviews to understand how parents of children with cancer manage their child's cancer pain at home. A section on research design describes the philosophical position of the thesis and purpose of mixed methods design. Patient and public involvement in this research is then described. An overview of research setting and the sample population precedes sections for each data collection method which outline the sample size, data collection tools, procedure, and data analysis used. Data integration methods, strategies to minimise threats to validity, and ethical issues are described to end this chapter.

Results are presented in Chapter 6 beginning with survey, then pain diary, and then interview. Survey and pain diary findings follow a similar pattern which begins with data quality and distribution followed by analysis of participant response rate and rich description of both samples. Survey and pain diary findings are then presented using descriptive and inferential statistics. Interview findings begin with a rich description of the sample which is followed by detailed description of the findings presented in model format and supported by frequent quotations.

Chapter 7 is comprised of three sections which provide the integration of results. Firstly, statistical integration of survey and pain diary results are provided. Secondly, meta-inference generation is conducted using matrices based around data collection methods. These matrices provided a mechanism for sorting large datasets into meaningful inferences which are used in the third step of integration, in which research questions form the basis of integration via joint display.

In Chapter 8, integration continues as results are discussed using contiguous approach through narrative. The joint display in Chapter 7 forms the basis of discussion of each research question using literature and theoretical frameworks. Following this, findings stemming from the integrative review in Chapter 3 are discussed in light of findings from this research, wider literature, and theoretical frameworks. Next, analysis of the implications of Theory of Planned Behaviour for this research, as well as implications for this research on the Theory of Planned Behaviour are presented. Final sections describe strengths and limitations of this research.

Chapter 9 concludes the thesis with a summary followed by description of how this research contributes new knowledge. Implications for clinical practice, future research, and policy are outlined. The dissemination strategy is provided as well as reflections from the researcher using reflexivity. Finally, concluding remarks close the thesis.

## **1.9 Summary**

This chapter provides readers with orientation to the thesis. Pain is a frequent symptom for children with cancer leading to negative physical and psychological consequences in the short- and long-term. Children with cancer are spending more time at home, leading to parents taking over the pain management role from HCPs. Government and national policy recognise the importance of parents in managing children's cancer pain at home. A description of my personal background and experiences which led to this doctoral work was provided. Thesis organisation is set out to orient readers to its structure and content. The next chapter provides a review of literature relating to pain experienced by children with cancer at home and parents' management of that pain.

## **Chapter 2. Background literature review**

### **2.1 Overview of background literature chapter**

This chapter provides an integration of two rapid reviews in a single report. Both reviews were conducted with a view to ascertaining whether there is sufficient evidence for the development of interventions to support parents of children with cancer managing their child's pain at home. The integrated background literature reviews asks three literature review questions: Is pain at home a problem for children with cancer and their families? What is the pain manifestation of children with cancer at home? What is parents' role in managing children's cancer pain at home?

Knowledge stemming from this review led to the aim and research questions for the research reported in Chapters 4-9 of this thesis. These are presented along with conclusions to close the chapter.

### **2.2 Rationale for rapid reviews**

As described in Chapter 1, children with cancer experience pain throughout the cancer trajectory regardless of their age, gender, or diagnosis. Pain has negative short- and long-term, psychological and physical consequences. Children with cancer are now spending more time at home being cared for by their parents, and yet evidence from postoperative literature suggests parents may not be able to adequately manage their children's pain at home (Sutters *et al.*, 2012; Zisk *et al.*, 2010). There is extensive evidence on children's cancer pain from inpatient settings (Hedén *et al.*, 2013; Miller *et al.*, 2011; Plummer *et al.*, 2017; Zernikow *et al.*, 2006, 2008), but evidence on children's cancer pain at home is lacking. In 2011, Fortier and colleagues issued a call for research targeting children's cancer pain at home with a focus on parents' pain management (Fortier *et al.*, 2011b). At that time, they concluded that development of interventions to support parents managing children's cancer pain at home was premature.

In 2013, as a precursor to doctoral studies, a rapid review was conducted by the student, along with a team of researchers (Twycross *et al.*, 2015b). This initial review aimed to explore sources, prevalence, and impact of pain on children with cancer and their families, as well as parents' experiences of managing pain at home and their attitudes towards pain and pain medication. In 2016, at the commencement of doctoral studies, an update to the original rapid review was conducted with a view to creating an integrated report of the most up-to-date research. This chapter contains the integration of both rapid reviews presented in a single report structured around the literature review questions presented in Table 2.1.



Table 2.1: Literature review questions

Literature review question
1. Is pain at home a problem for children with cancer and their families?
2. What is the pain manifestation of children with cancer at home?
3. What is parents' role in managing children's cancer pain at home?

### 2.3 Method and study selection

Method, study selection, data extraction, and quality appraisal for the 2013 review can be found in publication (Twycross *et al.*, 2015b). As with the 2013 review, the 2016 update followed a rapid review approach which aims to produce results in a faster timeframe by streamlining the traditional systematic review processes (Armitage and Keeble-Ramsay, 2009; Ganann *et al.*, 2010). Rapid reviews achieve this by limiting searches by year, database, language, or source and by one researcher doing parts of the review where resources are limited (Ganann *et al.*, 2010). Although shortened timeframes may introduce bias, rapid review methods are appropriate for contexts in which the aim is to synthesise evidence and contextualise empirical findings (Armitage and Keeble-Ramsay, 2009; Ganann *et al.*, 2010) so were suitable for this research.

Search time parameters were from October 2013 (the date of the 2013 review search) until February 2016. Search terms and databases matched the 2013 review (Table 2.2). The search was designed to be as broad and inclusive as possible to minimise chance of studies being missed. A comprehensive list of databases were selected which focused on nursing, medical and psychology research. Selected papers were subject to a quality appraisal process using Caldwell *et al.* 2011 tool which aims to highlight strengths and weaknesses of studies and aid weighting of studies in synthesis (Caldwell *et al.*, 2011). This tool can be utilised for qualitative and quantitative methods with a set of generic criteria for any type of methodology, and a set of criteria specific to each methodology.

In the 2016 search, a total of 1,203 papers were found, 200 of which were duplicates, leaving 1,003 papers. After reviewing titles, 884 papers were removed, leaving 119 papers. Abstracts of remaining papers were reviewed, and 103 papers discarded leaving 16 papers which were subject to full review. Fourteen of these were discarded leaving two publications relating to parents' management of children's cancer pain at home (Fortier *et al.*, 2014; Zhukovsky *et al.*, 2015). This selection process is displayed in Figure 2.1. The following three sections comprise findings from both literature reviews in a single report, as they relate to the literature review research questions (Table 2.1).

Table 2.2: Search terms and databases searched

Search terms
Child OR Children OR Paediatric OR Pediatric AND Cancer pain
Databases searched
CINAHL PubMed Web of Knowledge Medline PsychINFO PsychARTICLES AMED

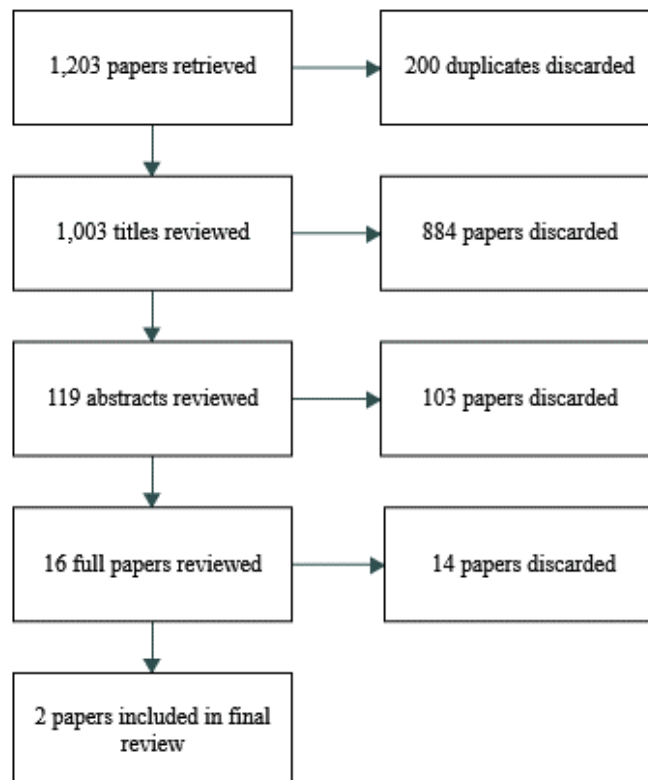


Figure 2.1: PRISMA flow diagram showing study selection from 2016 search

## 2.4 Is pain at home a problem for children with cancer and their families?

Results of both 2013 and 2016 literature combined provide a description of negative experiences of families of children with cancer due to cancer pain at home. Children's cancer pain can be a source

of anguish for families (Hedén *et al.*, 2013) which they describe as unendurable (Hellsten, 2000). At home, children's cancer pain was described as the worst type of emotional pain (Pöder *et al.*, 2010). Parents report feeling distressed (Hedén *et al.*, 2013) and frustrated (Hellsten, 2000) when seeing their child in pain. In interviews, parents of children with cancer described their child's pain as horrible, frightening pain which left them feeling helpless (Ferrell *et al.*, 1994a). Caring for children's symptoms was ranked as the highest level of burden associated with having a child with cancer by parents in a survey (Ferrell *et al.*, 1994b). These papers were published over 20 years ago and may not be reflective of current cancer treatments due to the changes outlined in Section 1.2.1. In addition, these papers did not specify whether pain referred to was inpatient or outpatient and the sample included only children who had recent pain experiences. It is unknown how these studies relate to current cancer treatments or to a sample of children who do not have recent cancer pain. More recently, parents of children with cancer asked to rate symptoms over the past week, regarded pain as one of the five most distressing symptoms at one week, two months, and four months following diagnosis (Pöder *et al.*, 2010). It is unknown whether this pain was experienced in hospital or at home. Children usually spend their first 10 days to two weeks after diagnosis in hospital, so it is likely that only the second and third time points refer to pain at home and even these may have included pain experienced during hospital admissions.

One study provided evidence of the problem of pain at home in children with cancer using a solely outpatient sample. In the US, children (n=55) aged 4-12 years, and their parents were asked to complete a two-week pain diary (Fortier *et al.*, 2014). Diary data were compared to baseline data including demographics, personality characteristics, parents' attitudes and misconceptions regarding analgesic drug use in children, and child QOL. Pain at home had the biggest hindrance on functioning in extracurricular activities, followed by household, social, sleep, and academic domains. Although applicability of home contexts is hindered by including samples with a mix of inpatient and outpatient children, combined evidence from 2013 and 2016 reviews, reveals that pain is a problem which is "clearly significant" (Twycross *et al.*, 2015b, p. 11) for children with cancer and their families at home.

## **2.5 What is the pain manifestation of children with cancer at home?**

Children's cancer pain manifestation at home is evidenced by a study carried out in Jordan which found 9/21 outpatient children had pain on the day of the study (Forgeron *et al.*, 2006). Location, duration, cause, and intensity of pain is unknown as the remainder of the study reports results from inpatient and outpatient participants together. A sample of parents of children both inpatient and outpatient rated pain as one of the five most common symptoms at one week, two months, and four months following diagnosis (Pöder *et al.*, 2010). Children with leukaemia and their parents

reported pain at each of seven time points in treatment trajectory (Van Cleve *et al.*, 2004). The most frequent pain locations were legs, followed by abdomen, head/neck and back. Due to inclusion of inpatient participants, the relevance of these studies to children's cancer pain at home is limited.

Four studies have used samples of outpatient children only to identify pain manifestation in children with cancer at home (Bossert *et al.*, 1996; Fortier *et al.*, 2014; Gedaly-Duff *et al.*, 2006; Zhukovsky *et al.*, 2015). In a US study, semi-structured interviews were used with children (n=20) aged 4-16 years, and their parents, to ascertain the location, source, and intensity of pain at home as well as the effectiveness of pain management strategies (Bossert *et al.*, 1996). Two children took part in a longitudinal arm of the study which involved keeping pain diaries. One child completed the whole pain diary, but the other child made only sporadic recordings. All children experienced pain which is perhaps unsurprising given that recent pain experience was an inclusion criterion. The most common locations were stomach, joints, legs, and back. Chemotherapy was identified as the most common cause of pain. Several limitations mean this research cannot accurately quantify pain experiences of children with cancer at home. Firstly, there is evidence that location of data collection (a hospital) may have confounded results with participants conflating their experiences at home and in clinical settings. Specifically, participants mentioned pain from procedures such as lumbar punctures which could not have happened outside healthcare settings. Secondly, due to the small sample size, no inferential statistics could be performed on either arm of this study. Thirdly, only two participants provided data on pain frequency so findings in this specific area are unlikely to be robust. Finally, management of children's cancer has changed significantly since this study was conducted over 20 years ago and findings may no longer be applicable. Despite limitations, this study presents emerging evidence of children with cancer experiencing pain at home.

In another US based study, diaries were used to collect data on several symptoms, including pain, from children aged 8-16 years and their parents at home (Gedaly-Duff *et al.*, 2006). This pilot study had a small sample (n=9) and study duration of just three days following administration of vincristine, a chemotherapy known to cause pain for children with cancer (Mora *et al.*, 2016). Pain was experienced by children throughout the three days. However, due to this study focusing on several symptoms, reporting of pain data were minimal. Data that are reported may be confounded by misunderstanding as additional information suggests pain reported may not have been cancer-related: for example, one patient reported pain due to a grazed knee from a cycling accident. Despite these issues, this study suggests children's pain manifestation continues at home following chemotherapy.

Zhukovsky (2015) and colleagues asked children aged 7-18 years with advanced cancer, and their parents, recruited from an outpatient cancer centre in the US, to separately rate pain over the previous 48-hour period alongside recording other symptoms. Pain was the most common symptom for children aged 10-18 years. This study provides only a retrospective measure of pain among other symptoms and recall bias may have confounded results. The short duration provides only a snapshot of children's pain presence or absence and does not address pain intensity, location, cause, or frequency. Regardless, this study provides evidence that pain is frequently present for children with cancer even at home.

Finally, Fortier and colleagues (2014) conducted a study which included retrospective reporting of pain for three months prior to study commencement and a two-week real-time data collection period using pain diaries. Children retrospectively reported having experienced chronic and recurrent pain, with the most common sites being back, legs and abdomen. This study benefited from a larger sample size (n=55). Although this study had longer duration of real-time data collection, it did not report on children's pain intensity and cause. The limited duration for real-time data collection (two weeks) may also not be sufficient to measure patterns in pain experience. Despite these limitations, this research provides real-time evidence of children with cancer having frequent pain at home, and retrospective evidence of chronic and recurrent pain at home.

In summary, although much evidence is limited by use of mixed inpatient and outpatient samples, four key studies (Bossert *et al.*, 1996; Fortier *et al.*, 2014; Gedaly-Duff *et al.*, 2006; Zhukovsky *et al.*, 2015) used outpatient only samples to quantify the pain manifestation of children with cancer at home. Each study suggested children with cancer experience pain at home. Due to methodological limitations including sample size and duration of data collection, three studies (Bossert *et al.*, 1996; Gedaly-Duff *et al.*, 2006; Zhukovsky *et al.*, 2015) were unable to quantify pain location, cause, intensity or frequency for children with cancer at home. The final study (Fortier *et al.*, 2014) had a larger sample and longer duration of data collection which enabled reporting of pain duration and location, but pain intensity and cause were not reported. In summary, no research has fully described the pain manifestation of children with cancer at home, and literature review question two remains unanswered.

## **2.6 What is parents' role in managing children's cancer pain at home?**

Literature relating to this research question is presented in two sections: parental pain assessment and parental pain management. Several studies report inadequacies in parental pain assessment (Ferrell *et al.*, 1994a, 1994b; Forgeron *et al.*, 2006; Fortier *et al.*, 2012). In interviews conducted

with 22 parents, 15 of whom represented children who were currently outpatients, parents reported they believed children should be responsible for expressing their own pain (Forgeron *et al.*, 2006). Misconceptions regarding pain expression were endorsed by a sample of parents representing children who were a mix of both inpatient and outpatient, on and off treatment at point of data collection (Fortier *et al.*, 2012). Neither of these studies used samples of only outpatients so it is difficult to ascertain the extent to which findings represent how parents assess their child's cancer pain at home. Nevertheless, this evidence indicates parents of children with cancer may be unprepared for their pain assessment role.

In terms of pain management, parents of children with cancer reported feeling unprepared for their pain management role which was a source of stress (Ferrell *et al.*, 1994a). Misconceptions regarding pain management have been reported which led authors to conclude parents lacked adequate pain management knowledge (Ferrell *et al.*, 1994b). These two studies were conducted over 20 years ago and may not represent current cancer treatment. In a quantitative survey, parents of children on and off treatment were found to have negative attitudes and misconceptions towards analgesic medications (Fortier *et al.*, 2012). Literature is conflicted regarding whether parents' use of pharmacological interventions increases (Lu *et al.*, 2011) or decreases over time (Van Cleve *et al.*, 2004). In a mixed methods study, of the 20 children who reported current pain, only seven received medications and seven received non-pharmacological interventions (Forgeron *et al.*, 2006). Although 21/35 children represented in this study were outpatient, it is unknown whether those who received interventions were at outpatient or inpatient. The qualitative element of this study involved interviews with parents. Authors concluded from interviews that parents lacked knowledge of the pharmacological interventions available to them at home. Parents in this study did not use opioids or non-pharmacological interventions at home. If their child was in pain, parents felt their only option was to go to hospital. This suggests adequate pain management could prevent hospital admissions. In a study of children with neuroblastoma, parents reported using non-pharmacological interventions (Lu *et al.*, 2011). Parents of children with leukaemia listed stressor modification which included sleep, hot/cold, and massage as well as pharmacological interventions, as their preferred choice of pain intervention (Van Cleve *et al.*, 2004). These studies are limited by samples including a combination of both inpatient and outpatient children. Reporting does not separate these children so the extent to which findings are relevant to understanding parents' management of children's cancer pain at home is unknown.

Two studies used a purely outpatient population (Bossert *et al.*, 1996; Fortier *et al.*, 2014). Bossert and colleagues (1996) collected a list of 35 different pain management strategies from children (n=20) and their parents at an outpatient clinic. Variations in the number of strategies used was associated with demographic variables suggesting that choice of pain management technique may

be influenced by family cultural background. However, there was no measure of frequency of use, effectiveness of each technique in response to pain, or reasons for choosing each technique. This evidence suggests there may be many different strategies for managing children's cancer pain at home.

Fortier and colleagues (2014) provide important detail on parents' pain management at home by investigating a sample of children who were, at point of investigation, cared for by their parents at home. Parents recorded analgesic administrations in response to child pain over a two-week period. On average, 2.5 children per day reported clinically significant pain of three or more on the numerical rating scale (NRS) but did not receive any analgesic drug interventions. Of the 55 children who participated, 60% did not receive an analgesic drug for the two-week duration of the study. Negative attitudes regarding avoidance of analgesic drugs, and misconceptions regarding the appropriate use of these drugs were correlated with fewer analgesic drug administrations. Parents most frequently cited not intervening because they did not think their child was in pain. Other reasons included a lack of prescribed pharmacological interventions, or the child refusing pharmacological interventions. Parents' misconceptions when measured quantitatively were significantly associated with not administering analgesic drugs. Real-time data collection negated risk of recall bias. This study did not allow parents to record interventions other than administration of analgesic drugs, meaning that parents who had administered non-pharmacological interventions would appear to have made no intervention. In addition, no measure of parents' pain assessment abilities was conducted, so it cannot be known whether inadequate pain assessment was the reason for lack of analgesic drug administration. Fortier and colleagues (2014) concluded that improvement in parents' pain management abilities would reduce children's pain at home. They concluded that gaps remained in several areas including: use of non-pharmacological interventions; exploration of barriers to pain management; and investigation of effective interventions to improve pain management (Fortier *et al.*, 2014).

The 2013 review concluded "no studies had investigated parents' actual pain management practices in this context" (Twycross *et al.*, 2015b, p. 12). Although Fortier and colleagues (2014) are able to address this to some extent, the absence of investigation of non-pharmacological interventions means parents' pain management practices are only partially represented. Research question three which asks what parents' role is in managing children cancer pain at home, remains unanswered.

## **2.7 Conclusions from background literature review**

Results of the background literature review have been presented concurrently in three sections corresponding to the three literature review questions (Table 2.1).

### ***2.7.1 Limitations of background literature***

None of the four key studies described above included children under four years of age (Bossert *et al.*, 1996; Fortier *et al.*, 2014; Gedaly-Duff *et al.*, 2006; Zhukovsky *et al.*, 2015). As children transition through cognitive developmental stages, their understanding and experience of pain changes (Craig, 1987). There is no indication that younger children experience any less pain, and some studies have found higher levels of pain in younger children with cancer (Friedrichsdorf *et al.*, 2007; Lautenbacher *et al.*, 2017). It is important that future research does not neglect further this already underrepresented group.

There is a good evidence base supporting physiological, sociocultural, provider, and systemic factors as potential causes for ethnic and cultural differences in pain experience, expression, and management (Campbell and Edwards, 2012). Each of the four key studies described above was conducted within the US. Although there are cultural similarities between the US and the UK, it is worth considering the differences before applying this literature to the UK, National Health Service (NHS) context.

On a sociocultural level, in children, it has been suggested that pain assessment, rather than pain experience may be the cause of cultural differences in pain (Finley *et al.*, 2009). Finnish and American parents have been found to hold different attitudes towards analgesic drugs (Kankkunen *et al.*, 2008). Similar findings are evident in a population of parents of children with cancer where ethnicity and language affected parents attitudes towards analgesic drugs (Fortier *et al.*, 2011a). Although these studies do not show differences between the UK and US specifically, evidence suggests cultural differences exist and that direct application of US studies to the UK may not be appropriate.

On a systemic, provider level, the US operates an entrepreneurial model of healthcare which means the competitive market is maximised and government involvement minimised (Brown, 2003). Healthcare is seen as a privilege and 16% of the population are uninsured (Roe and Liberman, 2007). Depending on state, US children's cancer services may not be free at point of service and even insured children may pay in part or have their healthcare limited depending on their individual



insurance package (American Cancer Society | Information and Resources about for Cancer, 2018). The American Society of Clinical Oncology report inequality in access to cancer services, especially services provided at home (American Society of Clinical Oncology, 2014). Conversely, the UK operates a market minimised, government maximised system, where healthcare is a right and coverage universal (Brown, 2003). Importance of healthcare equality is emphasised in NHS values which include “working together for patients”, and “everyone counts” (Department of Health and Social Care, 2015). These key differences between UK and US healthcare systems put them at opposing ends of the health system spectrum (Roe and Liberman, 2007).

As a result of the potential sociocultural and systemic differences, research conducted outside the US is needed. No substantive work has been conducted in a UK setting, and no assessment of the generalisability of these studies has been undertaken. Research in this thesis proposes to identify the relevance of previous studies to systems outside the US, specifically within the UK, NHS context.

### ***2.7.2 Implications for clinical practice***

Understanding how parents of children with cancer manage their child’s pain at home has important implications for nurses, parents, and children. Nurses play a key part in preparing parents for caring for their child at home (Bettle, 2015; Flury *et al.*, 2011). To empower parents in their pain management responsibilities at home, nurses need to know about the pain children are likely to experience at home as well as parents current state of pain assessment and pain management knowledge. Nurses will benefit from understanding the context in which parents manage pain at home and barriers they face in this task. Parental empowerment is likely to reduce distress and frustration faced by parents managing their child’s pain at home (Hedén *et al.*, 2013). In due course, effective pain management by parents of children with cancer at home, will reduce unnecessary pain experienced by these children and may prevent hospital admissions (Forgeron *et al.*, 2006).

### ***2.7.3 Summary of background literature***

Literature provides clear evidence in answer to literature review question one that pain is a problem for children with cancer and their families at home. Qualitative and quantitative evidence from many studies suggests that child cancer pain is a burden and hindrance to children and families at home. In answer to literature review question two, gaps remain regarding pain manifestation of children with cancer at home. Similarly, gaps in literature have been found regarding parents’ role in managing children’s cancer pain at home (literature review question three). There is little

information regarding how effectively parents are able to assess their child’s pain at home, what they do in response to their child’s pain, and what influences their choice of response. Table 2.3 summarises this report. In conclusion, the call for research targeted at parents’ management of children’s cancer pain at home (Fortier *et al.*, 2011b) remains unmet. There is a need for research to understand how parents of children with cancer manage their child’s pain at home.

Table 2.3: Summary of evidence for literature review questions

	<b>Literature review question</b>	<b>Summary of evidence</b>
<b>1</b>	Is pain at home a problem for children with cancer and their families?	Pain causes distress, anguish, burden, helplessness, frustration. Evidence of pain hindering several domains of life.
<b>2</b>	What is the pain manifestation of children with cancer at home?	Children do experience pain at home. Methodological limitations mean intensity, cause, and location are unknown.
<b>3</b>	What is parents’ role in managing children’s cancer pain at home?	Parents show misconceptions regarding pain assessment. Parents under-medicate children’s pain. Use of non-pharmacological techniques is unknown. Efficacy, frequency, and reasons parents choose strategies to manage pain is unknown.

## **2.8 Aim and research questions for this thesis**

The aim and research questions for the empirical research described in the remainder of this thesis are outlined in Table 2.4. The review of background literature above found that manifestation of children’s cancer pain at home is largely unknown leading to research question one. In addition, background literature found that information on how parents assess their child’s pain, what they do to manage their child’s pain, and what influences their choice of pain management interventions is incomplete. These findings led to research questions two, three and four respectively.

Table 2.4: Aim and research questions

<b>Aim</b>
To understand how parents of children with cancer manage their child’s cancer pain at home.
<b>Research questions</b>
1. What is the pain manifestation of children with cancer at home?
2. How do parents assess their child’s pain at home?
3. How do parents intervene to manage their child’s pain at home?
4. What influences parents’ choice of interventions to manage their child’s pain at home?

## 2.9 Summary

This chapter identified gaps in literature regarding parents’ management of children’s cancer pain at home. Research clearly identifies that pain at home causes distress for children with cancer and their families as well as hindering several domains of life. Little evidence exists describing the manifestation of children’s cancer pain at home. There is a scarcity of literature examining parents’ role in managing their child’s pain at home, with information on frequency and efficacy of non-pharmacological techniques incomplete. Some evidence suggests parents under-medicate their child’s cancer pain at home with analgesic drugs. Similarly, the interaction between parents’ pain assessment abilities, attitudes towards pain medication, and pain management practices is unknown. Finally, there is little information on barriers and facilitators to parents’ pain management in this context.

Of the four studies which use outpatient only samples and attempt to quantify children’s cancer pain at home (Bossert *et al.*, 1996; Fortier *et al.*, 2014; Gedaly-Duff *et al.*, 2006; Zhukovsky *et al.*, 2015), all have been conducted in the US. In addition, none of these studies investigate children under the age of four leaving a need to investigate this under-researched population. Filling the gaps in the literature identified in this review has implications for HCPs, parents and children with cancer whose pain the research presented in this thesis will ultimately attempt to minimise. Remaining chapters describe research which aims to understand how parents of children with cancer manage their child’s cancer pain at home. Once this research is conducted, researchers may consider developing interventions to support these parents. To enable effective intervention development, there is a need to identify interventions which currently exist to help parents manage their child’s pain at home and to ascertain which aspects increase likelihood of the intervention being successful. A literature review designed to address this is presented in the next chapter.

## **Chapter 3. An integrative review of interventions to support parents managing children's pain at home**

### **3.1 Overview of integrative review chapter**

This chapter provides an integrative review of interventions to support parents managing children's pain at home. Rationale for the review is provided followed by a description of methods used and attempts to increase reliability and validity. A descriptive summary of results is provided as well as analysis of potential reasons for intervention effectiveness and risk of bias within and across studies. Findings are discussed with limitations, practice implications, and research implications described.

### **3.2 Rationale for reviewing interventions to support parents managing children's pain at home**

Chapter 2 concluded that there was insufficient evidence for development of interventions to support parents managing children's cancer pain at home and that gaps remained regarding children's pain manifestation at home and parents' pain assessment and management role. Empirical research described in this thesis attempts to meet these gaps and increase understanding of parents' management of children's cancer pain at home. The development of interventions to support parents managing children's cancer pain at home is likely to follow this research. Intervention development is costly and involves several stages including design, piloting, evaluation, reporting, and implementation (Craig *et al.*, 2008). Careful planning is required to overcome practical and methodological challenges (Melnyk and Morrison-Beedy, 2012). Examining interventions and aspects which drive their effectiveness can inform future intervention development (Owen *et al.*, 2012). At time of review, no interventions existed to support parents of children with cancer in managing pain, but interventions did exist to support parents managing children's pain at home caused by other acute and chronic conditions. As part of the doctoral research presented in this thesis, an integrative review was conducted and subsequently published which examined interventions to support parents managing children's pain at home (Parker *et al.*, 2018). This chapter presents the findings of that integrative review.

A search of the Database of Abstracts of Reviews of Effects revealed two recent, relevant reviews (Chorney *et al.*, 2014; Kankkunen *et al.*, 2004). Both reviews restricted inclusion criteria to include postoperative literature only and consequently analysed a limited number of studies with small to moderate effect sizes found. Results of these two literature reviews were insufficient to understand the full range of factors that contributed to effective interventions to support parents' pain

management at home. No similar reviews were found in a search of The Cochrane Library. The review reported in this chapter broadened the search of previous reviews to encompass pain from other sources not limited to postoperative pain. Expanded inclusion criteria enabled full consideration of reasons for effectiveness or non-effectiveness of interventions by allowing comparison between greater number and type of interventions. Results included any intervention aimed at supporting parents managing children's pain at home, provided the pain was caused by an acute or long-term condition. A diverse range of study designs were included. Integrative review methods were utilised (Whittemore and Knafl, 2005) and narrative description was used to allow for studies with different methodologies to be represented (Bowman, 2007).

### **3.3 Methods**

#### **3.3.1 Objectives**

1. To identify interventions aimed at supporting parents when managing their child's pain at home.
2. To ascertain which aspects of interventions make them effective.

#### **3.3.2 Design**

This review followed Economic and Social Research Council guidance on narrative synthesis for systematic reviews (Popay *et al.*, 2006) and guidance for undertaking reviews in health care from the Centre for Reviews and Dissemination (CRD) (Akers *et al.*, 2009).

#### **3.3.3 Search strategy**

Search strategy development was an iterative process (Teddlie and Tashakkori, 2009) displayed in Table 3.1. Scoping searches from a selection of databases were conducted using four search strategies. Each strategy had a different level of specificity and sensitivity. A high sensitivity search would mean more papers would be selected incurring higher resource cost due to time spent screening papers but reducing the chance that relevant papers were missed. A high specificity search would reduce the number of papers selected which would reduce resources required to screen papers but increase chances of missing relevant papers. This combination of scoping searches helped to maximise sensitivity but increased specificity in line with resources.

Table 3.1: Potential search strategies derived using PICOT

Search One	Search Two	Search Three	Search Four
Child* OR pediatric OR paediatric OR adolescen* OR young adult* OR teenage*	Child* OR pediatric OR paediatric OR adolescen* OR young adult* OR teenage*	“parents’ pain management” OR “parent management of pain”	Child* OR pediatric OR paediatric OR adolescen* OR young adult* OR teenage*
AND	AND		AND
Parent* OR caregiver* OR guardian* OR famil*	Parent* OR caregiver* OR guardian* OR famil*		Parent* OR caregiver* OR guardian* OR famil*
AND	AND		AND
Pain management	Pain management		Pain management
	AND		AND
	Home OR “outside \$2 healthcare setting”		Limiters: English language publications, publications relating to humans, publications relating to children
			AND
			NOT PICU or “paediatric intensive care” OR “pediatric intensive care” OR death OR dying OR bereave OR “painful procedures” OR immunisation OR immunization OR inject* OR pregan* OR labour OR labor

Table 3.2 displays the numbers of papers from each scoping search. Search three did not reveal sufficient number of papers to give confidence that all relevant papers had been captured. Although search two provided greater numbers of results, the addition of the final criteria “home” OR “outside \$2 healthcare setting” might have meant relevant studies in which the recruitment strategy was within the healthcare setting were missed. Search one was selected to ensure maximum coverage. This was refined into search four by search limiters and Boolean operators to increase specificity in line with researcher resources but without limiting sensitivity. Limiters were inserted where possible and Boolean operators were applied to the title of papers.

Table 3.2: Results of initial scoping searches

	Search 1	Search 2	Search 3	Search 4
CINAHL*	1,674	174	3	1,244
Science direct	380	71	4	290
PubMed	384	46	3	297
Scopus	2,814	48	5	521
Web of Knowledge	802	233	5	618
Total	6,054	572	20	2,970

\*Includes: Medline, PsychINFO, PsychARTICLES, AMED

### 3.3.4 Eligibility criteria

Studies were assessed as eligible according to Population, Intervention, Comparison, Outcome and Time (PICOT) criteria (Liberati *et al.*, 2009) delineated in Table 3.3. All empirical methodologies were included. Papers published from the origin of the database until the date of the search (May 26<sup>th</sup>, 2016) were included. Due to resource limitations, only papers written in English were included and grey literature was excluded. Non-empirical publications, policy, and opinion papers were excluded as this review focused on empirical work. Reference lists of literature reviews were hand searched for relevant publications, but literature reviews were excluded as their inclusion could result in undue weight applied to studies included in both literature reviews and retrieved via search strategy.

Table 3.3: Inclusion criteria according to PICOT

Population			Intervention	Comparison	Outcomes			Time
Children	Parents	Cause of pain	Intervention	Comparison	Parents' pain management	Pain in children	Home	Time
Studies investigating children from birth to 18 or studies including teenagers and young adults included where there was evidence which assisted in meeting the objectives.	Studies investigating mothers, fathers, guardians or any individual with primary caregiving responsibility for the child.	Any disease or medical procedure which causes children to be in pain at home.	Intervention aims to reduce child pain or increase analgesic drug administration. AND Intervention aims to support parents at home.	Any comparison will be included.	Any aspect of parents' knowledge / attitudes towards pain management. OR Administration of analgesic drugs or non-pharmacological methods of pain management. OR Other aspects of pain management by parents. OR Parent assessment of child pain at home.	Self-report of pain by child. OR Proxy-report of pain by parents. OR Surrogate outcomes shown to be indicators of pain (e.g. heart rate). OR Any pain assessment tool including non-validated tools.	Any setting in which children's pain management is solely parents' responsibility	Any duration of data collection will be included.



### **3.3.5 Data sources and study selection**

Guidance from CRD (Akers *et al.*, 2009) was used for study selection. The following databases were searched from date of inception to 26<sup>th</sup> May 2016: MEDLINE; CINAHL Plus; PsychINFO; PsychArticles; AMED; PubMed; Scopus; Web of Knowledge. These databases were selected to provide maximum coverage of medical, nursing, psychology, alternative medicine, and scientific publications. Authors of potentially useful papers were contacted if the complete paper could not be obtained. Reference lists of included articles as well as literature reviews extracted from databases were hand searched to identify further studies.

### **3.3.6 Data collection and analysis**

Included papers were uploaded to NVivo™ (Version 10, QSR International) to aid review. CONSORT guidelines were used in deciding which items to extract from each study (Schulz *et al.*, 2010). The following items were extracted: aims, design, participants (number), participants (condition), intervention, intervention details, comparison, outcome measures, measure of child pain, duration of follow up, success in reducing child pain, success in increasing analgesic drug administration, success in other outcome measures, conclusions, reasons attributed to success/failure of intervention, comments. Statistical significance (using  $p \leq .05$ ) was used to judge success or failure of each intervention. Three summary measures were chosen: reduced child pain, increased analgesic drug administration, and reasons attributed to intervention success or failure. Whilst reduced child pain and increased analgesic drug administration may appear synonymous, some papers only reported one outcome measure, and other papers reported different findings between these two measures, so both were required summary measures. Reduction in child pain was considered a primary outcome measure and increased analgesic administration a secondary outcome measure. Reasons which authors of papers attributed to interventions success or failure was a summary measure which enabled this review to meet the objective of ascertaining which aspects of interventions make them effective.

Using the objectives as a focus (Borenstein *et al.*, 2009), an assessment of homogeneity in terms of participants, interventions, and outcomes contributed to deciding whether it was appropriate to conduct a meta-analysis (Haidich, 2010; Russo, 2007). Many interventions targeted children postoperatively, but meta-analysis would not have been possible due to heterogeneity in interventions and outcomes with differences in target, mechanism, and resources. Consequently a narrative synthesis was conducted using Economic and Social Research Council methods programme (Popay *et al.*, 2006) and CRD (Akers *et al.*, 2009) guidance. The reasons authors of included studies attributed to the success of interventions were analysed using methods similar to content analysis (Hsieh and Shannon, 2005). Alternative methods of qualitative analysis, for

example thematic analysis, were considered but it was concluded this was not appropriate as the text was not sufficiently rich or in-depth to provide themes (Braun and Clarke, 2006). Content analysis was chosen to ensure key information was derived from the text as a whole without over-weighting the importance of this data (Hsieh and Shannon, 2005). Text relating to why authors believed their intervention was effective or not was extracted and uploaded to NVivo™ (Version 10, QSR International). This text was then read repeatedly to provide a sense of data as a whole from which codes were derived, sorted into categories, and then subcategories. Exemplars for each subcategory were selected.

### **3.3.7 Quality appraisal**

#### **3.3.7.1 Risk of bias in individual studies**

The Critical Appraisal Skills Programme checklist was used to assess 13 randomised controlled trials (RCTs) (Singh, 2013). This checklist was chosen as it was developed by a group of experts, pilot tested, is frequently updated as required, and is used by many HCPs in research. The remaining studies were appraised using Caldwell and colleagues' framework which aims to highlight the strengths and weaknesses of each study. The framework was chosen as it includes specific criteria for qualitative and quantitative research, as well as a standard set of criteria for any methodology (Caldwell *et al.*, 2011). Using a structured risk of bias assessment enabled the narrative synthesis to put more emphasis on studies with minimal risk of bias (Akers *et al.*, 2009).

#### **3.3.7.2 Risk of bias across the dataset**

A funnel plot could not be constructed to ascertain publication bias due to the heterogeneity of studies. Instead, an examination of the significance of studies was conducted to ascertain whether studies may have been conducted but not published (Liberati *et al.*, 2009). In addition, a search for follow-up studies to included studies which were described as feasibility or pilot studies was conducted. This was achieved by searching for publications by authors of these studies. The purpose of this was to ascertain whether studies had been missed in the search strategy or conducted but not published.

## **3.4 Results**

### **3.4.1 Study selection**

The number of papers from each source was recorded and the selection process presented in a PRISMA flow diagram (Liberati *et al.*, 2009) (Figure 3.1). Of the 17 studies selected, 13 were RCTs, three quantitative, and one mixed method. Sample size ranged from 47-108 participants with

a median of 70 and interquartile range of 64.5. Two studies had no control group, and eight studies used standard care. Follow-up ranged from 3-109 days with a median of four days. Most studies (n=15, 88%) addressed postoperative pain. One study considered migraine pain and another chronic idiopathic pain.

### ***3.4.2 Descriptive summary***

Studies are described according to the target of the interventions within interactions between nurse, parent and child. Figure 3.2 shows three individuals present in parental management of children's pain at home: the nurse, the parent, and the child. Interventions vary in whether they target parents directly, interactions between the nurse and parent, or interactions between the parent and child. Multifaceted interventions target more than one interaction. Characteristics of included studies and key points from risk of bias assessment are available in Appendix 1.

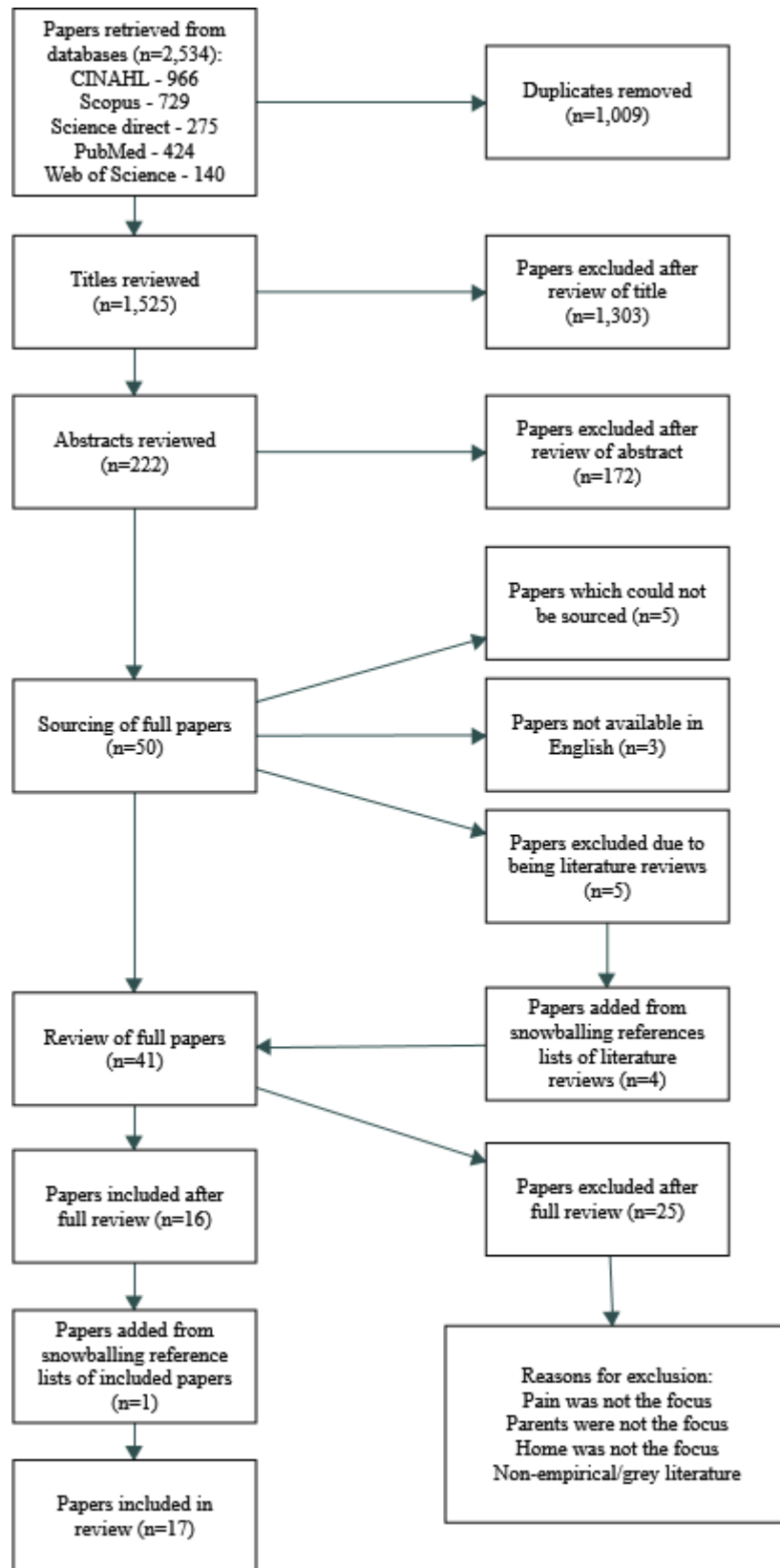


Figure 3.1: Search results and study selection

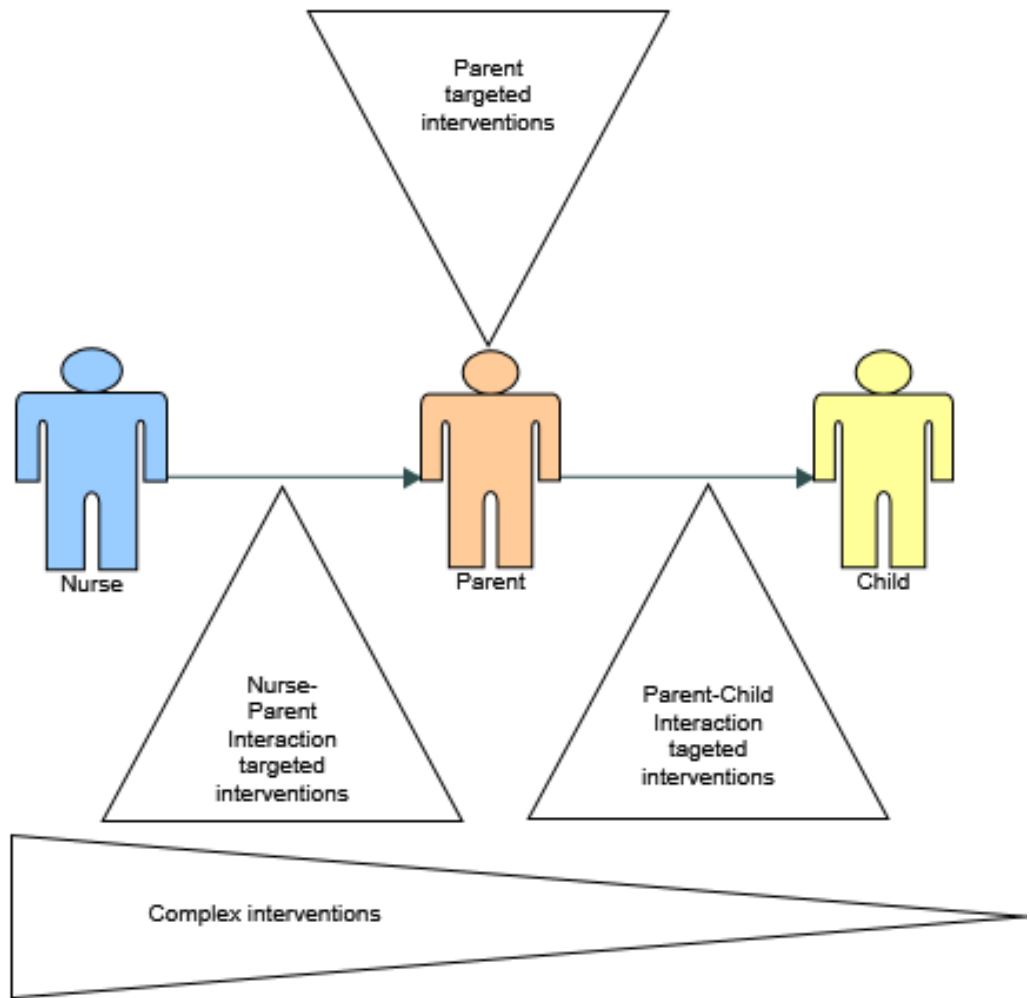


Figure 3.2: Classification of interventions according to intervention target

### 3.4.2.1 Parent-targeted interventions

Three RCTs addressing postoperative pain (Sutters *et al.*, 2004, 2010; Wiggins, 2009) encouraged parents to administer analgesic drugs regularly rather than when required. This type of intervention is termed “around-the-clock” (ATC). Wiggins and colleagues (2009) as well as Sutters and colleagues (2004), found an ATC intervention statistically significantly increased analgesic drugs received by children, but no statistically significant reduction in pain was observed. Sutters and colleagues (2004) concluded this was due to inadequate strength of the analgesic drug administered (paracetamol with codeine). Sutters and colleagues repeated their intervention in 2010 with paracetamol and hydrocodone finding a statistically significant reduction in pain scores at specific time points.

Four RCTs (Allen and Shriver, 1998; Bailey *et al.*, 2015; Chambers *et al.*, 1997; Helgadóttir and Wilson, 2014) and one quasi-experimental study (Vincent *et al.*, 2012) used parent education interventions. Apart from Allen and Shriver (1998) who addressed migraine pain, these

interventions all addressed postoperative pain. Two interventions provided written information only (Bailey *et al.*, 2015; Chambers *et al.*, 1997), the remainder provided written and verbal information. Bailey and colleagues (2015) found oxycodone information increased parents' satisfaction and significantly reduced pain scores at two of the three time points. Helgadóttir and Wilson (2014) found statistically significantly lower pain behaviour (a measure of how the child's behaviour differs from their usual behaviour) when distraction and pain management education were provided compared with pain management education alone. Analgesic drug administration was not measured and there was no significant pain reduction. No significance in pain reduction or analgesic drug administration were reported by Vincent and colleagues (2012), although there was a significant increase in child satisfaction. Chambers and colleagues' (1997) pain education booklet did not reduce child pain despite significantly altered attitudes and increased analgesic drug administration. A combination of biofeedback and parent education resulted in increased adaptive functioning and significant pain reduction compared to a control group receiving biofeedback alone (Allen and Shriver, 1998).

#### **3.4.2.2 Nurse-parent interaction targeted interventions**

Two studies targeted nurse-parent interactions (Paquette *et al.*, 2013; Sepponen *et al.*, 1999). Telephone calls by nurses following surgery did not reduce pain but did statistically significantly increase analgesic drug administration (Paquette *et al.*, 2013). Despite statistically significantly increasing analgesic drugs administered by parents and influencing analgesic drug choices, an education programme provided to doctors and nurses did not reduce pain (Sepponen *et al.*, 1999).

#### **3.4.2.3 Child-parent interaction targeted interventions**

Pain assessment was considered an interaction between child and parent and as such, pain assessment tools were considered to be targeting child-parent interactions. None of the three RCTs which provided pain assessment tools to parents found a statistically significant reduction in child pain or increase in analgesic drug administration (Franck *et al.*, 2007; Kankkunen *et al.*, 2009; Unsworth *et al.*, 2007). Provision of instructions for the Wong-Baker Faces Pain Scale (WBFPS) (Wong and Baker, 1988) and analgesic drug administration increased administration of codeine as instructed and reduced unnecessary drug administration (Unsworth *et al.*, 2007). Providing children with a temporary tattoo of the WBFPS made no difference to pain or analgesic drug administration when compared to a paper version (Franck *et al.*, 2007). Provision of the Parents Postoperative Pain Measure made no difference to problems faced by parents managing postoperative pain (Kankkunen *et al.*, 2009).

Chronic idiopathic pain was addressed through a family-directed, internet-based, cognitive behavioural therapy (CBT) RCT delivered online (Palermo *et al.*, 2009). Skills such as deep breathing and muscle relaxation were covered in specific child and adult modules. Compared to waitlist controls this intervention resulted in statistically significant reductions in pain and activity limitations. An RCT found administering take-home analgesic drugs did not have any effect on pain or analgesic drug administration compared to parent supplied analgesic drugs (Hegarty *et al.*, 2013).

#### **3.4.2.4 Multifaceted interventions**

Two papers addressed postoperative pain using a combination of techniques (Sutters *et al.*, 2012; Walther-Larsen *et al.*, 2016). One mixed methods study used written information, follow-up phone calls, nurse coaching, and provision of a timer (Sutters *et al.*, 2012). One prospective observational cohort study used verbal parent education, tailored provision of analgesic drugs, and a pain assessment tool (Walther-Larsen *et al.*, 2016). Both studies concluded the intervention resulted in reduced pain but neither used a control group.

#### **3.4.3 Reasons for intervention success or failure**

Three categories arose from an analysis of the reasons authors of the studies attributed to the success or failure of their interventions (Table 3.4): characteristics of interventions; components of parents' pain management which were addressed by interventions; and key features of the research investigating intervention efficacy. Two subcategories of characteristics of interventions were identified: multifaceted interventions and tailored interventions. Authors of seven papers stated either that the intervention failed due to addressing only one factor, or attributed its success to multifaceted design (Franck *et al.*, 2007; Hegarty *et al.*, 2013; Kankkunen *et al.*, 2009; Sutters *et al.*, 2010, 2012; Vincent *et al.*, 2012; Walther-Larsen *et al.*, 2016). In three papers, authors stated that tailoring their intervention to the patient was key in intervention effectiveness (Chambers *et al.*, 1997; Sutters *et al.*, 2010; Walther-Larsen *et al.*, 2016).

Table 3.4: Reasons authors attributed to the success or failure of interventions

Category	Subcategory	No. of references	Exemplars
Characteristics of the intervention	Multifaceted intervention	7	"Instead of addressing only one of many barriers to effective pain management following day surgery in children, we decided to implement as many interventions as feasible." (Walther-Larson <i>et al.</i> , 2015)
	Tailored	3	"A limitation of this study is that the ... booklet used in this study was a general booklet ... booklet more specific to day surgeries, with step-by-step instructions for postoperative pain management, may be even more effective..." (Chambers <i>et al.</i> , 1997)
Components of parents' pain management	Analgesic drug effectiveness	8	"Development of more optimal analgesic agents is needed to lower pain intensity." (Paquette <i>et al.</i> , 2013)
	Pain education	7	"...the need for additional education for home pain and symptom management that provides knowledge about interventions that can be implemented from the time of discharge through the lengthy recovery." (Wiggins <i>et al.</i> , 2009)
	Pain assessment tools	2	"In future studies of pain assessment ... could turn out to be a valuable tool, both in research and clinical care." (Walther-Larson <i>et al.</i> , 2016)
	Attitudes	2	"This study also provides some preliminary evidence indicating that both parents' attitudes toward children's pain medications and how they assess their children's pain contribute independently to how they medicate their children's pain." (Chambers <i>et al.</i> , 1997)
Key features of the research	Adequate sample size	7	"... it was underpowered, which could result in a failure to observe a difference when in truth there was one." (Franck <i>et al.</i> , 2007).
	Measure of adherence	2	"... one limitation of this investigation is the absence of a direct measure of parental compliance with implementation of the guidelines." (Allen & Shriver, 1998)
	Measure of pain behaviour	1	"Pain behaviour or overall pain may capture the effects of the intervention better than pain intensity." (Helgadottir & Wilson, 2014)
	Measure of sedation	1	"The sedative properties of acetaminophen and hydrocodone may have affected the interpretation of behavioural observations and contributed to lower FLACC scores." (Sutters <i>et al.</i> , 2012).

Components of parents' pain management were grouped into four subcategories: analgesic drug effectiveness, pain education, pain assessments, and attitudes. Success or failure of interventions was attributed to effective or ineffective analgesic drugs the in eight studies (Bailey *et al.*, 2015;



Helgadóttir and Wilson, 2014; Paquette *et al.*, 2013; Sepponen *et al.*, 1999; Sutters *et al.*, 2004, 2010; Walther-Larsen *et al.*, 2016; Wiggins, 2009). Success or failure of interventions was attributed to parent education by authors of seven studies (Chambers *et al.*, 1997; Sutters *et al.*, 2004, 2010, 2012; Vincent *et al.*, 2012; Walther-Larsen *et al.*, 2016; Wiggins, 2009). Brief education could be ineffective, but overloading information could also be detrimental (Vincent *et al.*, 2012). Most effective information was tailored to the situation (Chambers *et al.*, 1997) and written information alone could be sufficient (Sutters *et al.*, 2004, 2010). Intervention success was attributed to pain assessment tools by authors of two papers (Chambers *et al.*, 1997; Walther-Larsen *et al.*, 2016). Parents' attitudes towards pain management including misconceptions regarding tolerance, side-effects, and pain expression were considered by authors of two papers to be a barrier to intervention effectiveness (Chambers *et al.*, 1997; Vincent *et al.*, 2012).

Features of the research were grouped into four subcategories: adequate sample size, measure of adherence, measure of pain behaviour, and measure of sedation. Inadequate sample size was considered a potential cause of intervention failure by eight authors (Allen and Shriver, 1998; Franck *et al.*, 2007; Kankkunen *et al.*, 2009; Paquette *et al.*, 2013; Sutters *et al.*, 2010; Unsworth *et al.*, 2007; Vincent *et al.*, 2012; Walther-Larsen *et al.*, 2016). Two interventions were considered ineffective due to non-adherence (Allen and Shriver, 1998; Chambers *et al.*, 1997) although one had an indirect measure of adherence (Allen and Shriver, 1998). In one study, a measure of pain behaviour provided pain assessment over a longer period rather than a snapshot in time (Helgadóttir and Wilson, 2014). This study found pain behaviour improved statistically significantly and authors attributed the success of their intervention to using this pain assessment tool. A sedation measure was suggested by one author as a feature of research which would ensure sedation is not mistakenly measured as low pain (Sutters *et al.*, 2012). Two studies assessed sedation as an intervention outcome (Sutters *et al.*, 2010, 2012). In one of these studies, sedation increased as pain decreased, suggesting the pain reduction may have been a product of increased sedation rather than true pain management (Sutters *et al.*, 2012).

#### **3.4.4 Quality appraisal**

Key points from the risk of bias assessment can be found in Appendix 1. All studies included risk of bias due to methodological limitations: nine studies were underpowered (Allen and Shriver, 1998; Chambers *et al.*, 1997; Franck *et al.*, 2007; Kankkunen *et al.*, 2009; Paquette *et al.*, 2013; Sutters *et al.*, 2010; Unsworth *et al.*, 2007; Vincent *et al.*, 2012; Wiggins, 2009); pre-intervention group differences existed in five studies (Bailey *et al.*, 2015; Chambers *et al.*, 1997; Franck *et al.*, 2007; Paquette *et al.*, 2013; Sutters *et al.*, 2010); inappropriate randomisation was evident in four studies (Hegarty *et al.*, 2013; Helgadóttir and Wilson, 2014; Kankkunen *et al.*, 2009; Palermo *et*

*al.*, 2009); three studies provided no information on characteristics of participants who withdrew (Hegarty *et al.*, 2013; Kankkunen *et al.*, 2009; Sutters *et al.*, 2004); two studies did not report how participants were randomised (Allen and Shriver, 1998; Chambers *et al.*, 1997); no information on group homogeneity was given in two studies (Hegarty *et al.*, 2013; Wiggins, 2009); and two studies lacked a control group (Sutters *et al.*, 2012; Walther-Larsen *et al.*, 2016).

Findings that only two studies lacked any statistically significant outcome in at least one variable suggests there may be bias against publication of non-significant studies (Dwan *et al.*, 2013). No follow-up study could be found for three of the studies reported as feasibility or pilot studies (Franck *et al.*, 2007; Sutters *et al.*, 2012; Wiggins, 2009) suggesting these studies are either unpublished or not conducted. The search strategy revealed two potentially useful papers which could not be located. Abstracts alone cannot ascertain whether retrieving these papers may have influenced this review.

### **3.5 Discussion**

Interventions to support parents managing their child's pain at home have been evaluated in this review, which considers the primary outcome of reducing child pain and secondary outcome of increasing analgesic drug administration. Excluding interventions which lacked a control group, the group (Figure 3.2) which produced the highest number of interventions successful in reducing child pain was parent-targeted interventions. Of the eight studies in this category, three (two parent education and one ATC) led to a statistically significant reduction in child pain. Interventions targeting parents directly and those targeting nurse-parent interactions were most likely to be successful in the secondary outcome measure of increasing analgesic drug administration. All nurse-parent interaction interventions (the telephone follow-up and the doctor and nurse education) and half of the parent-targeted interventions (all three ATC and one of the three parent education) statistically significantly increased analgesic drug administration.

In total, six interventions were successful at increasing analgesic drug administration but only three were successful at reducing child pain. This suggests an increase in analgesic drug administration does not guarantee a reduction in child pain. It may be easier for an intervention to statistically significantly increase analgesic drug administration than reduce child pain. One reason for this may be inadequacies in analgesic drugs used (Bailey *et al.*, 2015; Helgadóttir and Wilson, 2014; Paquette *et al.*, 2013; Sutters *et al.*, 2004, 2010; Walther-Larsen *et al.*, 2016; Wiggins, 2009). When Sutters and colleagues changed from paracetamol and codeine (2004) to paracetamol and hydrocodone (2010), a previously ineffective intervention became successful in reducing child

pain. It is of note that codeine is no longer recommended for use in children (MHRA, 2013). There is evidence that genetic differences in metabolism may hinder its effectiveness which may have been a reason for the lower efficacy of interventions which used codeine (Department of Health, 2013; Van Hout *et al.*, 2014).

Another reason for the lack of interventions which effectively reduce children's pain may be inaccurate pain assessment. Most studies assessed pain intensity on a scale. Studies which used other pain-related measures including pain behaviour (Helgadóttir and Wilson, 2014), activity limitations (Palermo *et al.*, 2009), and satisfaction with pain levels (Vincent *et al.*, 2012), found interventions were effective at influencing these. Historically self-report has been considered the gold-standard for children's pain assessment but recently recognition of other individual and contextual factors has been advised (Twycross *et al.*, 2015c). These alternative measures provide a measure of pain over time and may be a way of measuring individual and contextual factors.

Although both multifaceted interventions lacked a control group, both claimed success in reducing child pain. Seven authors listed the multifaceted nature of interventions as a characteristic which increased intervention success. Frameworks are available to guide researchers in the development of multifaceted interventions (Campbell and Edwards, 2012; Craig *et al.*, 2008) for which there is a growing recognition (Campbell *et al.*, 2000). This review and area of research is limited by the high risk of bias within studies primarily due to being underpowered. Significant differences may not have been detected where they existed due to small sample sizes (Schulz and Grimes, 2005).

Chorney and colleagues' review concluded that ATC interventions were most effective at improving parents' management of postoperative pain at home, but that interventions involving parent education and pain assessment provision were ineffective (2014). This review concurs with the efficacy of ATC interventions but adds in support of parent educational interventions which have been found successful in improving analgesic drug administration. This may be particularly relevant where ineffective analgesic drugs have been the cause of ineffective interventions. Chorney and colleagues (2014) grouped interventions according to mechanism of action rather than intervention target. This grouping, in addition to their inclusion of only postoperative interventions which led to a smaller dataset being reviewed, meant Chorney and colleagues conclusions diverged from this review.

### **3.5.1 Methodological limitations of literature review**

Due to limited resources, non-English publications were excluded, Boolean operators and limiters were applied in the search strategy, and grey literature could not be included. This may have limited the search and meant relevant articles may not have been detected but searching reference lists of included articles was conducted to minimise risk of missing articles. Areas of concern in every study identified by the risk of bias assessment limited meaningful inferences (Caldwell *et al.*, 2011; Singh, 2013). Awareness of these areas enabled more accurate conclusions to be drawn (Akers *et al.*, 2009). Many interventions may no longer be relevant as 6/17 included articles were published more than 10 years ago.

Trustworthiness and credibility should be considered when interpreting reasons authors attributed to the success or failure of interventions (Cope, 2014; Noble and Smith, 2015). This content analysis was conducted by one researcher in isolation with no peer debriefing, triangulation, or negative case analysis (Hsieh and Shannon, 2005). Due to study heterogeneity, meta-analysis could not be conducted, instead a robust narrative synthesis was conducted with the same goal: to collate the dataset and draw conclusions (Popay *et al.*, 2006). The recognition for narrative synthesis is growing and even when meta-analysis is possible, at times narrative synthesis is recommended (Centre for Reviews and Dissemination, 2008).

It is possible that studies which aimed to support parents managing children's pain at home may have increased use of non-pharmacological interventions whilst having no effect on pharmacological interventions. The decision not to include use of non-pharmacological interventions as an outcome measure in this integrative review may mean such findings were not detected. Appendix 1 provides a table of characteristics of studies selected for the integrative review. This table includes a summary of "other outcomes" investigated in these studies. Examination of these outcomes reveals a lack non-pharmacological outcome investigations which suggests these were not routinely measured in studies which investigated interventions to support parents managing children's pain at home. Future reviews may warrant consideration of these interventions in efficacy considerations.

### **3.5.2 Implications**

This integrative review suggests parent-targeted interventions were most likely to be successful at reducing child pain at home which in turn suggests it is appropriate for research in this thesis to focus on parents and use parents as participants. Multifaceted interventions may be more likely to be effective. There may be multiple aspects to parents' management of children's pain at home

which may be best investigated using multiple data collection techniques and mixed methods. The design of research in this thesis has taken into account findings that parents' attitudes and misconceptions may affect their pain management at home in two ways: an assessment of parents' attitudes was conducted during data collection; and attitudes are a key component of the theoretical framework, which was used to guide research in this thesis.

Findings of the review described in this chapter help bridge the gap in literature between the investigation of research in this thesis and intervention development. Researchers implementing this review in intervention development will increase the likelihood of producing effective interventions. Interventions to assist parents managing their child's cancer pain at home should: target parents directly or target nurse-parent interactions; be tailored and multifaceted; consider analgesic drug effectiveness, pain education, and pain assessment. Studies investigating these interventions will be more likely to show true efficacy if they have adequate sample size and consider measures of adherence, pain behaviour, and sedation. A discussion of these findings in light of the investigation of research described in this thesis and theoretical frameworks will be conducted in Chapter 8, Section 8.6.

### **3.6 Summary**

This chapter presents evidence from a published integrative review using narrative synthesis to identify interventions which aimed to help parents manage their child's pain at home and ascertain which aspects of interventions make them effective (Parker *et al.*, 2018). Parent-targeted interventions were most effective at reducing child pain at home, but many interventions may have been limited by ineffective analgesic drugs. Analgesic drug administration was most effectively increased by parent-targeted interventions and interventions targeting nurse-parent interactions. Key features of research, components of parents' pain management, and characteristics of interventions which would increase intervention effectiveness were discussed. Non-pharmacological interventions were not considered as an outcome measure for this study but may be important in future reviews. Implications of this review on research in this thesis and beyond were described. The next chapter describes how theoretical frameworks have been used in research described in the remainder of this thesis.

## Chapter 4. Theoretical framework

### 4.1 Overview of theoretical framework chapter

This chapter describes how theoretical frameworks have been selected and used in this research. Initially, preliminary searches for a theoretical framework are described. Following this, two theoretical frameworks are described in detail. Evidence for each theoretical framework is presented and rationale for theoretical framework selection provided. Finally, explanation of how The Theory of Planned Behaviour has been used in this research is outlined.

### 4.2 Preliminary searches

Use of theoretical frameworks will increase understanding of theory and research has found that interventions for children with long-term conditions are more effective if they have a clear theoretical underpinning (Aldiss *et al.*, 2015). Use of theoretical frameworks in research should be explicit (Michie *et al.*, 2009). Criteria used in searching for appropriate theoretical frameworks include simplicity, predictability, testability, internal consistency, and coherence to other theories (Agnew & Pyke, 1969).

The integrative review in Chapter 3 revealed that parent targeted interventions were most likely to be successful in reducing child pain at home. Background literature (Chapter 2) suggests more understanding of parental behaviour in managing pain for children at home is required. As a result, a preliminary search was conducted into both parenting and behavioural theories. The search of parenting theories revealed several theories focusing on the impact of parenting styles on child development (Ainsworth, 1990; Baumrind, 1967; Bowlby, 2005). Child development is not the focus of this research, rather this study is interested understanding parent behaviour and how to effectively modify that behaviour so focus of the search moved to behavioural theories.

Preliminary searching for behavioural theories revealed numerous theoretical frameworks including:

- Lippitt's Phases of Change Theory (Senft, 1960);
- Risk-Resilience Model (Wallander *et al.*, 1989);
- Lewin's Three-Step Change Theory (Kritsonis, 2005);
- Health Belief Model (Hochbaum *et al.*, 1952);
- Diffusion of Innovation Theory (Rogers, 1976);

- Transtheoretical Model (Prochaska and Velicer, 1997); and
- Social Norms Theory (Elster, 1989).

A multidisciplinary team tasked with creating guidelines for behaviour change interventions in health conducted a search of psychological literature and found that although such literature was extensive, no adequate theory existed (Abraham *et al.*, 2009; NICE, 2007). Two key theories were heavily cited in this guidance and many key psychological concepts identified as important were drawn from these theories. These two theories will now be considered in greater depth.

### **4.3 Theory of Planned Behaviour**

The Theory of Planned Behaviour (TPB) is depicted in Figure 4.1. It was proposed by Ajzen in 1985 as an extension to the Theory of Reasoned Action (TRA) which states that behaviour is a result of intention, which is in turn the result of two key concepts: attitudes and subjective norms (Fishbein, 1979). Attitudes result from interactions between behavioural beliefs (what individuals believe will result from behaviour) and outcome evaluations (the extent to which individuals value the outcome). Subjective norms result from interactions between normative beliefs (how individuals believe others value a behaviour) and motivations to comply (the extent to which individuals want to comply). A key limitation of TRA is its weak link between intention and behaviour (DiClemente *et al.*, 2018). Addition of perceived behavioural control (PBC) strengthened this link by considering the extent to which external factors may inhibit or facilitate behavioural intent (Ajzen, 1985). Perceived behavioural control results from interactions between control beliefs (the extent to which individuals believe they can control behaviour) and control frequency (barriers to behaviour). Ajzen notes that PBC can also exert a direct effect on behaviour. This research focuses on TPB rather than TRA due to the inclusion of PBC, which incorporates barriers and facilitators likely to be influential in parental management of children's cancer pain at home as well as its most extensive use in relevant literature.

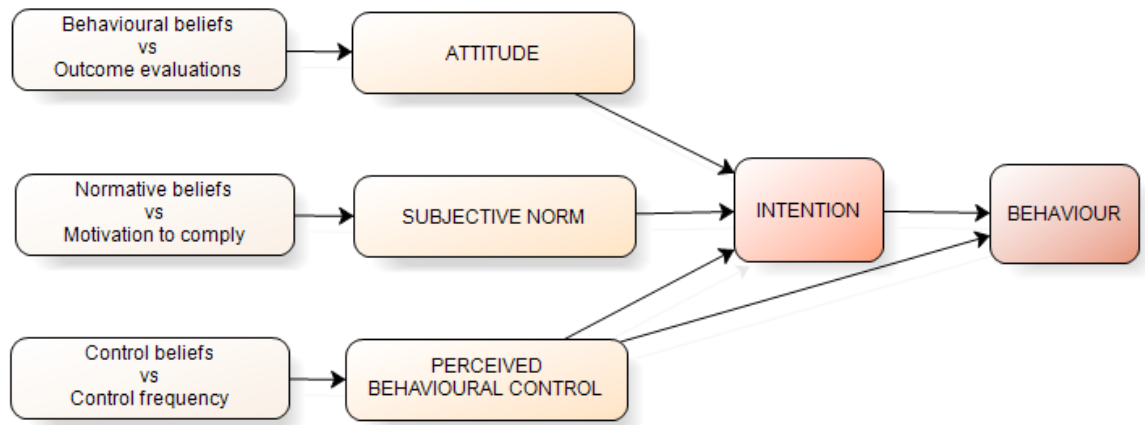


Figure 4.1: Visual representation of the Theory of Planned Behaviour

#### 4.4 Social Cognitive Theory

The Social Cognitive Theory (SCT) (Figure 4.2) posits that behaviour is a product of interactions between personal, behavioural and environmental factors (Bandura, 1986). Personal factors are cognisant of outcome expectations (the individual’s anticipated consequences), outcome expectancies (the value individuals place on consequences), and efficacy expectations (individuals’ confidence in their ability to perform) (Bandura, 2011). The concept of self-efficacy is closely related to PBC (Ajzen, 1991) but it is important to note the terms are not equal (DiClemente *et al.*, 2018). Where self-efficacy refers to the confidence an individual places in their ability to carry out an action, PBC relates to the extent to which an individual believes an outcome can be achieved with their own efforts. Environmental factors are a result of observational learning and reinforcement. Behavioural factors stem from observation (the way individuals perceive themselves), self-judgement (a comparison of the individual to others) and self-reaction (the conclusion individuals make about themselves) (Zimmerman, 1986).



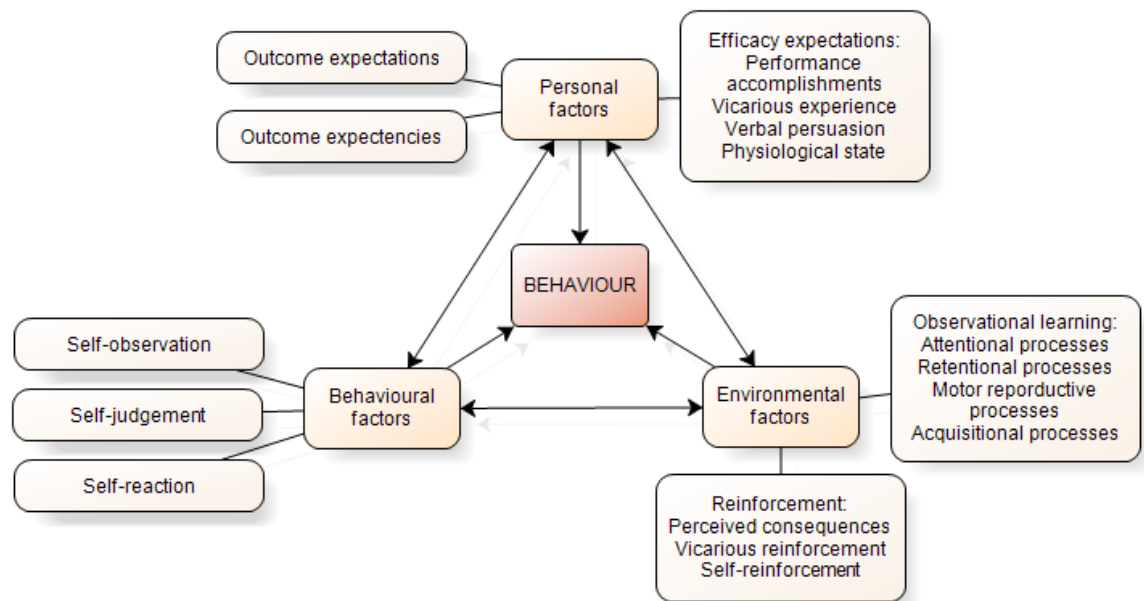


Figure 4.2: Visual representation of the Social Cognitive Theory

#### 4.5 Evidence for theoretical frameworks in health-related behaviour change

A systematic review found 21 interventions had used TPB to varying extents but only one addressed all components (Hardeman *et al.*, 2002). Positive changes were resultant from 42% of studies in which TPB was used for intervention development. The TPB has been an effective predictor of health related behaviour both within (Nash *et al.*, 1993) and outside (Janke, 1994) healthcare settings. It was utilised to design a survey investigating pain management intentions in nurses (Edwards *et al.*, 2001). Edwards and colleagues measured intention, rather than behaviour, but found TPB could explain 40% of the variance.

The SCT was used to design and implement a programme aimed at encouraging older adults with heart disease towards self-regulation (Clark *et al.*, 1992). The programme involved educational meetings, videotape, and workbook and group discussion, with the goal of increasing self-regulation and developing strategies to modify actions. There were areas of improvement attributed to the intervention; specifically self-efficacy was a significant predictor of disease management (Clark and Dodge, 1999). One study comparing these theories, found SCT to be a better predictor of attendance at a fitness centre than TPB (Jekauc *et al.*, 2015).

#### **4.6 Applicability of theoretical frameworks to health-related behaviour change**

Both theoretical frameworks have evidence to support their use in health contexts. The Social Cognitive Theory emphasises observation in learning, the role played by real or symbolic models, the environment, motivation, and past experiences. It has been suggested that SCT would provide a good theoretical framework for situations where the population are motivated to change (Hardeman *et al.*, 2002). There is evidence that parents are motivated to improve their pain management abilities (Lu *et al.*, 2011). However, although detail in a model can be a strength, SCT has been criticised for being too complex and comprehensive to be applicable to research (Anfara Jr and Mertz, 2014). In addition, the role of cognition and control as emphasised by TPB in learning is less evident in the SCT. The TPB is well suited to behaviours where individuals do not have complete control over their ability to perform (Ajzen, 2005). A key advantage of TPB is that its simplicity lends itself to empirical testing (Anfara Jr and Mertz, 2014) by enabling assessment of concepts and relationships within the theory (McMillan and Schumacher, 2010). For this reason, TPB was chosen to aid design, data collection, and analysis.

#### **4.7 Application of Theory of Planned Behaviour to research questions**

The TPB was used to guide selection and design of research methods. In analysis, the way in which TPB was applied varied for each research question (Table 2.4). Particular emphasis was given to how TPB can provide insight into behavioural elements of understanding parental management of children's cancer pain at home. This meant TPB was applied to how parents assess their child's pain at home and what influences parents' choice of interventions to manage their child's pain at home (research questions two and four). No direct application of TPB was made to children's cancer pain manifestation at home (research question one) as this is not related to parent behaviour. Similarly, TPB was not applied to how parents intervene to manage their child's pain at home (research question three) as the behavioural aspects of this question were addressed in research question four. Question two cannot be answered without the findings of question one. Similarly, question four cannot be answered without the findings of question three. In this way, TPB relates to all research questions although it is explicitly applied only to two. Not only did this research attempt to use TPB to provide insight into research findings but it additionally used research findings to provide insight to TPB thereby increasing theoretical knowledge.

## **4.8 Summary**

This chapter described the search for theoretical frameworks, which led to in-depth description and consideration of TPB and SCT. Evidence for each of these theoretical frameworks and rationale for how the TPB was selected is presented. Finally, the way in which TPB applies to each research question is given. The next chapter will describe methods used to understand how parents of children with cancer manage their child's cancer pain at home.

## Chapter 5. Methods

### 5.1 Overview of methods chapter

This chapter provides readers with an understanding of methods used in this research with rationale provided for decisions made. Opening sections provide the methodological approach, philosophical position and purpose of the mixed methods design. A description of the overall sampling strategy is provided, as well as the patient and public involvement (PPI) strategy. Research setting and sample are described followed by presentation of each data collection method (survey, pain diary, and interview) in a uniform structure which considers sample, data collection tools, procedure, and data analysis. In the final sections, data integration procedures are described followed by strategies for minimising threats to validity and lastly ethical issues are discussed.

### 5.2 Research design

Neither quantitative nor qualitative aspects could provide a complete understanding of research questions developed through analysis of gaps in the literature so a mixed methods approach was adopted (Dures *et al.*, 2011). A convergent parallel, mixed methods design was used (Creswell and Plano Clark, 2011). Data collection for all three methods were conducted in parallel over a period of six months. Table 5.1 shows how each data collection method contributed to each research question. Surveys were used to measure parental pain assessment and attitudes towards analgesic medications (research questions 2 and 4). Pain diaries were used to measure pain manifestation, provide reference material for discussion of parental pain assessment, measure parents' choice of interventions to manage pain, and record comments regarding what influences choice of interventions (research questions 1-4). Interviews were used to provide understanding of children's pain manifestation, parental pain assessment, choice of interventions, and the influencers of choice of interventions in managing children's pain at home (research questions 1-4). This study was designed to meet gaps in literature informed by the TPB which predicts that behaviour is influenced by intention, which is in turn influenced by attitudes, subjective norms and PBC (Ajzen, 1985). Attitudes were investigated using surveys and interviews. Subjective norms and PBC were investigated using interviews.

Table 5.1: Contribution of data collection methods to each research question

<b>Data collection method</b>	<b>Research question answered</b>	<b>How data answer research questions</b>
Surveys	2	Measured parents' misconceptions regarding pain assessment.
	4	Measured parents' attitudes towards analgesic drugs which influence choice of interventions to manage their child's pain.
Pain diaries	1	Measured baseline pain manifestation and provided reference material for discussion of this research question during interviews.
	2	Provided reference material for discussion of this research question in interviews.
	3	Measured parents' choices, frequency, and efficacy of interventions to manage children's pain at home.
	4	Parent comments contributed to understanding of what influences parents' choices of interventions.
Interviews	1	Understanding children's pain manifestation at home.
	2	Understanding how parents assessed their child's pain at home.
	3	Understanding what parents did to manage their child's pain at home.
	4	Understanding parents' decision-making process around pain management interventions.

### ***5.2.1 Philosophical position***

To fully meet the aim and answer the research questions, both qualitative and quantitative methods are required. This section describes the philosophical implications of combining quantitative and qualitative research. The paradigms associated with each type of research are described and philosophical difficulties in combining quantitative and qualitative methods are acknowledged. Pragmatism is presented as a paradigm for uniting ontological and epistemological barriers to produce philosophical justification for mixed methods research.

Many paradigms exist, but positivist and interpretivist paradigms will be discussed as these are most strongly associated with quantitative and qualitative research respectively. Many dualisms exist between these paradigms (O'Cathain, 2006). Quantitative research is associated with a positivist paradigm (Creswell and Miller, 2000). Positivists believe in a single, objective, and measurable reality (Feilzer, 2010). Epistemological assumptions stemming from this purport that knowledge can be gained via objective, reductionist, and deductive methodologies (O'Cathain, 2006), in which the observer is impersonal, passive and separate from research (Johnson and

Onwuegbuzie, 2004). Positivist methods are closed-ended techniques such as questionnaires and experiments which produce numerical data (O’Cathain, 2006).

Qualitative research is associated with the interpretivist paradigm sometimes termed the constructivist paradigm (Pope and Mays, 2006). Interpretivists reject a single reality in favour of multiple realities constructed socially (Guba, 1990). Epistemological assumptions stemming from this purport that knowledge can be gained subjectively, holistically, and inductively, resulting in qualitative methodologies (O’Cathain, 2006). The researcher is acknowledged as part of data generation and multiple truths are accepted (O’Cathain, 2006). Interpretive methods such as interviews, focus groups, and ethnography collect open-ended, word-based data (O’Cathain, 2006).

These opposing paradigms have led to what has been termed the “incompatibility thesis” (Howe, 1988; Johnson and Onwuegbuzie, 2004). Some scholars suggest qualitative and quantitative methods cannot and should not be mixed due to opposing epistemological and ontological standpoints (Greene and Caracelli, 1997; Guba, 1990; O’Cathain, 2006). To proceed with this research, a philosophical unification of qualitative and quantitative methods was required. Pragmatism, which is often used in mixed methods research (Creswell and Plano Clark, 2011; Johnson and Onwuegbuzie, 2004), offers a practical approach to this philosophical dilemma (Johnson and Onwuegbuzie, 2004). Using the pragmatic paradigm, it has been suggested that qualitative and quantitative methods are compatible (Howe, 1988). The compatibility thesis highlights similarities between quantitative and qualitative research and purports that combining paradigms is not only allowed, but often required for effective research (Howe, 1988). Pragmatism simultaneously recognises single and multiple realities (Creswell and Plano Clark, 2011) which are both naturally occurring and constructed (Reed *et al.*, 2016). Pragmatists emphasise similarities between positivist and interpretivist paradigms and consequently value both qualitative and quantitative research (Dewey, 1929; Johnson and Onwuegbuzie, 2004). Dualisms of positivist and interpretivist paradigms are rejected by pragmatists in preference for common-sense approaches (Johnson and Onwuegbuzie, 2004).

Positioning this research within the pragmatic paradigm means the importance of the research question is elevated above that of philosophical incompatibilities (Teddlie and Tashakkori, 2009). This epistemological justification allows use of the most appropriate methods of data collection for the aim and research questions. The pragmatic paradigm has led to the search for utility in methods selected, data collected, and analytical approach (Feilzer, 2010). Mixed methods has been recognised as a distinct research methodology (Greene, 2008). In research discussed in this thesis,

the pragmatic ontological perspective, warranted investigation of pain manifestation, pain assessment, pain management interventions and influences of intervention choice. Neither quantitative nor qualitative methods alone could adequately investigate these, and mixed methods was required. Pragmaticism allows combination of these methods within a single study with the purpose of producing results which are more than the sum of their parts (Teddlie and Tashakkori, 2009).

### ***5.2.2 Purpose of mixed methods design***

This study exhibits both inter-method mixing due to the use of more than one method of data collection, as well as intra-method mixing due to the use of both qualitative and quantitative components of the pain diary (Johnson and Turner, 2003). This research used mixed methods for the primary purpose of complementarity: by utilising both quantitative and qualitative methods, this study produced enriched understanding of parents' management of children's cancer pain at home, which would not have been obtained had a single method been used (Bryman, 2006; Dickson *et al.*, 2011; Greene *et al.*, 1989). After considering several typologies of purposes of mixed methods (Bryman, 2006; Creswell and Plano Clark, 2011; Teddlie and Tashakkori, 2009), Greene and colleagues' seminal paper from 1989 was chosen to guide description and implications of complementarity as the primary purpose for mixed methods. Complementarity is described as one of five purposes for mixed methods designs and influences seven design characteristics: methods, phenomena, paradigms, status, implementation independence, implementation timing, and study (Greene *et al.*, 1989). Each of these design characteristics will now be considered, being explicit about the way in which this study design will produce complementarity.

The extent to which methods used in the study differ from each other is referred to as the methods characteristic. Methods used in this study (quantitative survey, mixed methods pain diary with both numerical and free text responses, and qualitative semi-structured interviews) are considered different from one another in terms of the methodological paradigm from which they are derived. Weaknesses in quantitative methodology were countered by strengths in qualitative methodology and vice versa (O'Cathain and Thomas, 2006; Onwuegbuzie and Johnson, 2006). Quantitative surveys facilitate large samples which, due to time and resource limitations, would not be possible using qualitative interviews. Qualitative interviews facilitate richness and depth in data collection which could not be achieved with quantitative surveys.

Greene and colleagues state that complementarity can be achieved using methods which examine "*different facets of a single phenomenon*" (Greene *et al.*, 1989, p. 266). The single phenomenon is

parents' management of children's cancer pain at home. Different facets of this phenomenon include: pain manifestation (as measured via pain diaries and described in interviews); pain assessment (as measured via surveys and described in interview); pain management interventions (as recorded in pain diaries and described in interviews); and reasons for pain intervention decisions (as measured via surveys and described in interviews as well as pain diary comments). Complementarity is achieved in the investigation of these different facets of parents' management of children's cancer pain at home.

Complementarity is achieved through use of a single paradigm for all methods which are given equal weight (Greene *et al.*, 1989). In terms of implementation, Greene and colleagues stated complementarity designs should not be implemented independently but should be interacting and should be implemented simultaneously. Interaction of methods is found in three aspects of this study design: Firstly, interaction of sample – a subset of the same participants is used for each data collection method; Secondly, interaction in data collection – the pain diary was used as part of the interview to guide questions; Thirdly, interaction in data integration through use of statistical techniques, matrices, joint display, and the contiguous approach achieved through narrative. The final design characteristic states that complementarity designs should be a single study (Greene *et al.*, 1989). This research is considered one study and is presented as such.

This research did not seek triangulation as a purpose for using mixed methods. Several authors advocate caution in use of the term triangulation due to its wide application often without clear definition (Moran-Ellis *et al.*, 2006; O'Cathain, 2006; O'Cathain and Thomas, 2006; Teddlie and Tashakkori, 2009). In the classic sense, triangulation is the process of seeking convergence to increase validity (O'Cathain and Thomas, 2006; Petros, 2011; Teddlie and Tashakkori, 2009). This assumes methods are investigating the exact same phenomena which was not what this study aimed to do (Fetters *et al.*, 2013). This also assumes if methods produce convergent results that the methods do not have the same flaws, which cannot be guaranteed (Moran-Ellis *et al.*, 2006; O'Cathain and Thomas, 2006).

Greene and colleagues (1989) stated that their work was not exhaustive and anticipated additions to their list of purposes for mixed methods research. In addition to complementarity, this research used mixed methods for the secondary purposes of completeness, offset and explanation. In this research, mixed methods will not achieve completeness in the fullest sense, but rather will be used to achieve a more comprehensive understanding of the phenomenon (Bryman, 2006). Using a mixed methods sampling strategy which included both convenience sampling to increase breadth of



the sample, and purposive sampling to increase depth of the sample, meant the strengths and weaknesses of methods were able to offset one another (Bryman, 2006; Petros, 2011). It has been noted that it may not be possible to predict the outcomes of mixed methods research due to the vastness of the datasets generated. Consequently, new purposes for mixing may arise as the study progresses. In this research, although not originally the purpose of mixing, explanation emerged as a further purpose for mixing methods due to divergences in findings (Bryman, 2006).

### ***5.2.3 Overall sampling strategy and order of data collection***

Having a mixed methods sampling strategy which creatively combines quantitative and qualitative sampling techniques is a defining characteristic of mixed methods research (Teddlie and Yu, 2007). Sampling was based on the research questions and followed guidance from Creswell and Plano Clark (2011) for convergent designs and Teddlie and Tashakkori (2009) for parallel mixed methods sampling. Sampling for each data collection method was dictated by the aim and research questions and designed to maximise statistical benefits gained from a large quantitative sample and depth of information gained from a small qualitative sample (Creswell and Plano Clark, 2011). The mixed methods sampling strategy aimed to “*generate complementary databases that include information that has both depth and breadth regarding the phenomenon under study*” (Teddlie and Yu, 2007, p. 85). As previously mentioned, this is one way in which methods used in this research offset one another and produce complementarity (Bryman, 2006; Petros, 2011).

Convenience sampling from the population was used to recruit survey participants (procedure described in Section 5.5.3). Convenience sampling from the survey sample was used to recruit pain diary participants (procedure described in section 5.6.3). Use of convenience sampling aimed to increase breadth of sample (Parahoo, 2014). Purposive sampling of participants from the pain diary sample was used to recruit interview participants (procedure described in section 5.7.3). Use of purposive sampling aimed to increase depth of sample (Heavey, 2014; Parahoo, 2014). This constitutes concurrent mixed methods sampling (Teddlie and Yu, 2007): Sampling strategies for each data collection method were independent from each other, linked only by the use of a subset of survey sample for pain diary and a subset of pain diary sample for interviews.

Keeping the participant pool constant meant results from one data collection method could be legitimately related to results of another on an individual and collective level (Creswell and Plano Clark, 2011). This sampling strategy also meant that although this design is described as parallel, it also contains sequential aspects. Data were collected in parallel with surveys, pain diaries and interviews being completed by different participants simultaneously. However, each individual

participant transitioned through each data collection method in a uniform order. The order of data collection was designed to aid recruitment and facilitate data collection. Completion of survey and pain diary prior to interview removed the risk of the Hawthorne effect – that partaking in an interview will effect a participants’ response to future data collection methods (Todd, 2010). Participants completed the survey first, which aided recruitment in two ways. Firstly, the survey required the least participant time and was perceived to be easy (McKenna *et al.*, 2010), so participants who had reduced time capacity or desire to invest in the research were empowered to participate with minimal burden. Secondly, there are many ways researchers can design their study to increase participation (Morton, 2008). It is possible that completion of the survey may have enabled participants to understand more about the research and increased their interest and thus participation in pain diary and interview. Participants were given the option of completing the survey and taking part in an interview without completing a pain diary. However, all the interview participants did actually complete a pain diary. Participants completed the pain diary before the interview, which facilitated data collection by allowing the researcher to use and reference pain diaries during interviews. This facilitated discussion and allowed participants to expand on their pain diary responses.

### **5.3 Patient and public involvement strategy**

Patient and public involvement in research is increasingly recognised as important (Brett *et al.*, 2014). The Health Research Authority (HRA) defines PPI as research “*undertaken ‘with’ or ‘by’ patients and the public rather than ‘to’, ‘about’ or ‘for’ them*” (Buckland, 2007). Benefits of PPI have been evidenced at every stage of research and include better prioritisation of research topics, increased relevance of research materials to participants (Brett *et al.*, 2014), and increased rigour and impact of research (Involve, 2013). Challenges to PPI include role confusion, divergences of opinion, and tokenism (Brett *et al.*, 2014). Recommendations to counter these challenges include having clearly defined roles and responsibilities (Health Research Authority, 2016), building rapport (Brett *et al.*, 2014; Involve, 2013), and ensuring involvement is both transparent and influential (Legare *et al.*, 2011). This research aimed to maximise benefits whilst minimising challenges, to move beyond tokenism, and involve experts by experience<sup>2</sup> in as many stages of research as feasible. The PPI strategy described below provides a transparent outline of recruitment of experts by experience and roles undertaken.

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<sup>2</sup> For the purpose of this document, individuals who chose to participate in PPI will be referred to as “experts by experience” (McLaughlin, 2009)

Experts by experience were recruited through a variety of mechanisms. Parents attending a support group at The Royal Marsden NHS Foundation Trust were asked if they would be willing to be contacted by a researcher regarding an opportunity to collaborate on a research project. Those who expressed interest were sent an email. A children's cancer charity advertised this opportunity on their website and emailed potential experts by experience. An email was sent to the PPI group within The Royal Marsden NHS Foundation Trust.

Experts by experience were offered collaboration at differing levels of involvement which considered their preferences. Collaboration was depicted in terms of a ladder of involvement which allowed experts by experience to select which "rung" suited their current time capacity, other commitments, and desire to collaborate. Levels do not relate to the power afforded to experts by experience (Arnstein, 1969), but rather to a level of commitment chosen by each individual. At each level, interaction and opportunity to build rapport increased. This model was created out of a desire to facilitate involvement of every individual who wished to contribute regardless of capability and availability (Tritter and McCallum, 2006). Initial contact involved an email, which provided a brief research summary and role description for each level. To ensure experts by experience had accurate expectations of their involvement, these descriptions were transparent about the influence of their involvement and lack of monetary remuneration (Health Research Authority, 2016).

At the lowest rung of the ladder of involvement, experts by experience corresponded via email. Documents were sent via email and experts by experience were given opportunity to respond and discuss electronically. The middle rung involved the same collaboration as at the lowest rung, but in addition included telephone or Skype conversations between the researcher and expert by experience. Experts by experience were provided with a verbal description of the study and each document. The highest rung involved the same collaboration as at the lowest and middle rungs but in addition included a face-to-face meeting with the researcher. This could be a one-to-one meeting or a discussion group with several experts by experience. Higher levels of involvement provided more knowledge about experts by experience. At the conclusion of each interaction, experts by experience were asked if they would be happy to be contacted in future phases. Eight experts by experience contributed and their input was incorporated into several areas of this research. Prior to data collection, experts by experience were asked to comment on research design and documents including consent forms, participant information sheets, survey, pain diary (electronic and paper), and interview schedule. Prior to the completion of analysis, experts by experience were asked to comment on a summary of results from each data collection method. Throughout research experts by experience were sent biannual newsletters updating them on the status of the research.

## 5.4 Research setting and sample

Children's cancer services within the UK are organised such that children are allocated one tertiary cancer centre which provides initial and specialist care, and one shared care centre which is geographically more local and provides more immediate but less specialist care (NHS England, 2017). Participants were recruited from The Royal Marsden Hospital NHS Foundation Trust, a tertiary cancer treatment hospital within London, UK. Parents in this sample represented children whose care was managed by a range of shared care centres. Participants were recruited from two wards: the children's inpatient ward and the children's day unit. The children's ward is an 18-bed ward, which provides care for children from age 1-15 years old. Treatments offered include chemotherapy, radiotherapy, phase I and II clinical trials, and stem cell transplant. The day unit has 22 beds/chairs and 7 cubicles. Annually it provides care for more than 5,000 outpatient children and young people aged 1-24 years.

Participants were mothers, fathers, guardians or any individual with primary caregiving responsibility for a child aged from birth until the day before their 17th birthday with any cancer diagnosis, on treatment with curative intent. As children with cancer transition to adolescence and young adulthood, they begin taking greater role in their own self-management (Svedberg *et al.*, 2016). Inclusion criteria was age limited due to the aim of focusing on parents' role in pain management. Background literature revealed no differences in pain experienced by children regardless of their specific cancer diagnosis (Olson and Amari, 2015) so children were included with any cancer diagnosis. A historical trend towards treatment being the primary cause of pain for children with cancer has been found in background literature (Twycross *et al.*, 2015b), so the decision was made to include children only if they were on treatment with curative intent. Provided children were receiving treatment there was no minimum or maximum time since diagnosis. Children were excluded if their treatment was not curative as pain manifestation and management differs in this stage of treatment (Roza *et al.*, 2014). Parents were excluded if they were under the age of 18 or had insufficient English language and literacy to complete survey, pain diary or interview. Eligibility information was provided by HCPs who knew the family and acted as intermediaries. Healthcare professionals acted as gatekeepers and advised whether family circumstances, such as proximity to diagnosis, could mean participation caused additional distress (Creswell and Plano Clark, 2011; Teddlie and Tashakkori, 2009).

## **5.5 Survey method**

Table 5.1 shows how surveys were designed to answer research questions two and four. This research used surveys to measure attitudes of parents towards children's pain assessment, and analgesic drug interventions. Surveys are appropriate for assessing stable phenomena due to measuring only a snapshot in time (McKenna *et al.*, 2010). Attitudes have been shown to be relatively stable (Ajzen, 2006; Edwards *et al.*, 2007) so in this case, surveys were appropriate. Surveys are the most common method of attitude measurement (Ajzen, 2005) and have several advantages. Firstly, surveys are self-administered, which allows participants to choose the time and location of participation. Secondly surveys facilitate a large sample of participants to be recruited with relatively little cost (Parahoo, 2006). Finally, completion of surveys in the researchers' absence minimises social desirability bias (Jones and Rattray, 2010).

### **5.5.1 Sample**

Participants were recruited based on availability at time of recruitment rather than demographic criteria or representativeness. Although ideal sampling for minimising bias would be using a frame to select a random sample (Parahoo, 2014; Teddlie and Tashakkori, 2009), this would be unlikely to yield a sufficient sample size given study resources, timeframe, and attrition. For pragmatic reasons, a convenience sample was chosen to maximise recruitment. This research is primarily descriptive meaning that the aim of statistical analysis is to measure various parameters of interest and use these measurements to make inferences about the population. These inferences are made using the actual value and the confidence interval. A sample aim of 100 participants was chosen for pragmatic reasons: This would allow 95% confidence intervals for endpoints calculated with maximum width +/- 10% to be calculated with the exact method. A judgement was made in collaboration with a statistician that 10% width was sufficiently precise to be pragmatically useful in this research.

### **5.5.2 Data collection tools**

Two data collection tools, the Parental Pain Expression Perceptions questionnaire (PPEP) (Zisk *et al.*, 2010), and Medication Attitudes Questionnaire (MAQ) (Forward *et al.*, 1996) were used in combination. Question two was answered using the PPEP which assesses parents' attitudes towards pain expression in children (Zisk *et al.*, 2010). Question four was answered using the MAQ which was developed to measure mothers' attitudes toward paracetamol and opioid medication administration (Forward *et al.*, 1996). Both questionnaires consist of multiple items which increases reliability by reducing the impact of mistakes (Ajzen, 2005).

The PPEP was developed by a group of experts using relevant research, literature and clinical experience to produce nine items (Zisk *et al.*, 2007a). Each item is rated on a seven-point Likert scale with higher scores indicating greater attitudinal barriers. Internal consistency has been demonstrated in parents of children undergoing surgery with Cronbach's alpha at 0.79 (Zisk *et al.*, 2007a) and 0.78 (Zisk *et al.*, 2010). A previous factor analysis resulted in three sub-scales: active loud; quiet inactive; and attention seeking (Zisk *et al.*, 2007a). This scale was originally developed in the US for postoperative pain in children but has been used in the UK context with postoperative pain (Twycross *et al.*, 2014) and in the US with parents of children with cancer (Fortier *et al.*, 2014).

Item generation from researcher expertise and literature, with subsequent review by six paediatric pain researchers led to the development of the MAQ (Forward *et al.*, 1996). It was tested on a sample of mothers of healthy children and reliability and factor analysis conducted. A 16-item questionnaire with four sub-scales (addiction, side-effects, tolerance, and drug abuse) was constructed. Each item is rated on a seven-point Likert scale. In mothers of healthy children this scale had Cronbach's alpha of 0.68 (Forward *et al.*, 1996) and reached 0.73 when tested on parents of children undergoing surgery (Chambers *et al.*, 1997). Originally, four sub-scales existed for this scale, but a previous factor analysis resulted in this being reduced to three: avoidance; appropriate use; and fear of side-effects which are used in this thesis. This scale has also been utilised in the UK for children with postoperative pain (Twycross *et al.*, 2014) and in the US for children with cancer (Fortier *et al.*, 2012, 2014).

During the design phase, one expert by experience commented on statements in the MAQ such as "Children who take pain medication for pain may learn to take drugs to solve other problems" and "Using pain medication for children's pain leads to later drug abuse". This parent felt that seeing these statements, in the research context, would have increased her worries for her child and may have caused her to withhold pain medication. These concerns presented an ethical and methodological dilemma. Alterations to the MAQ could not be conducted without initiating a revalidation process and there are no suitable alternative questionnaires. The MAQ remained the preferred measurement tool. In response to this comment and as an attempt to counter ethical and methodological issues, the following statement was inserted at the end of the survey: "*If you have any questions or points for clarification please feel free to contact me*". The expert by experience who presented these concerns was contacted to explain the process and result of their comment.

Demographic data including parent age, gender, ethnicity, household income, and educational background as well as child age, gender, diagnosis, and time since diagnosis were collected. This enabled rich sample description which aided assessment of transferability and clinical applicability as well as assessment of sample representativeness. Demographic data were collected at the end to reduce the effect of participant fatigue on survey responses (Gerrard *et al.*, 2011). Survey length was minimised and instructions simplified to reduce participant burden and maximise completion rates (Parahoo, 2006).

### **5.5.3 Procedure**

Figure 5.1 outlines steps from initial potential participant contact, survey participation, and progression to subsequent data collection methods. Prior to data collection, the researcher met key members of clinical teams to increase awareness of the research and inform staff of its likely impact on them and their patients. Recruitment took place during daytime working hours approximately three days per week for a period of six months. Every patient was assessed for eligibility using inclusion and exclusion criteria with the help of HCPs. Healthcare professionals ascertained from eligible potential participants whether they were happy to be approached by a researcher. Potential participants who indicated they were happy to be approached were provided with written and verbal information by the researcher (Appendix 2). If requested, an hour later the researcher returned to provide an additional opportunity for potential participants to ask questions. If participants expressed interest, they were given the survey to complete at a time of their choosing, so that they did not need to return to the site to receive it (Appendix 3). Consent was implied by participants completing and returning the survey. Identifiable data were not collected during the survey unless participants indicated they would like to participate in future data collection methods. In this case, participants completed a “Please contact me” form attached to the survey. For these participants anonymity could not be maintained, but confidentiality was maintained for all participants throughout this research.

To test question clarity, completion time (Teddlie and Tashakkori, 2009), and acceptability of materials used (Jones and Rattray, 2010), a pilot test was conducted with three participants. Recruitment and procedures for this pilot stage followed those of the main study and gave participants an opportunity to provide feedback. This feedback may have resulted in changes of protocol prior to final study commencement. No changes were made as a result of the pilot, so pilot data were included in results.

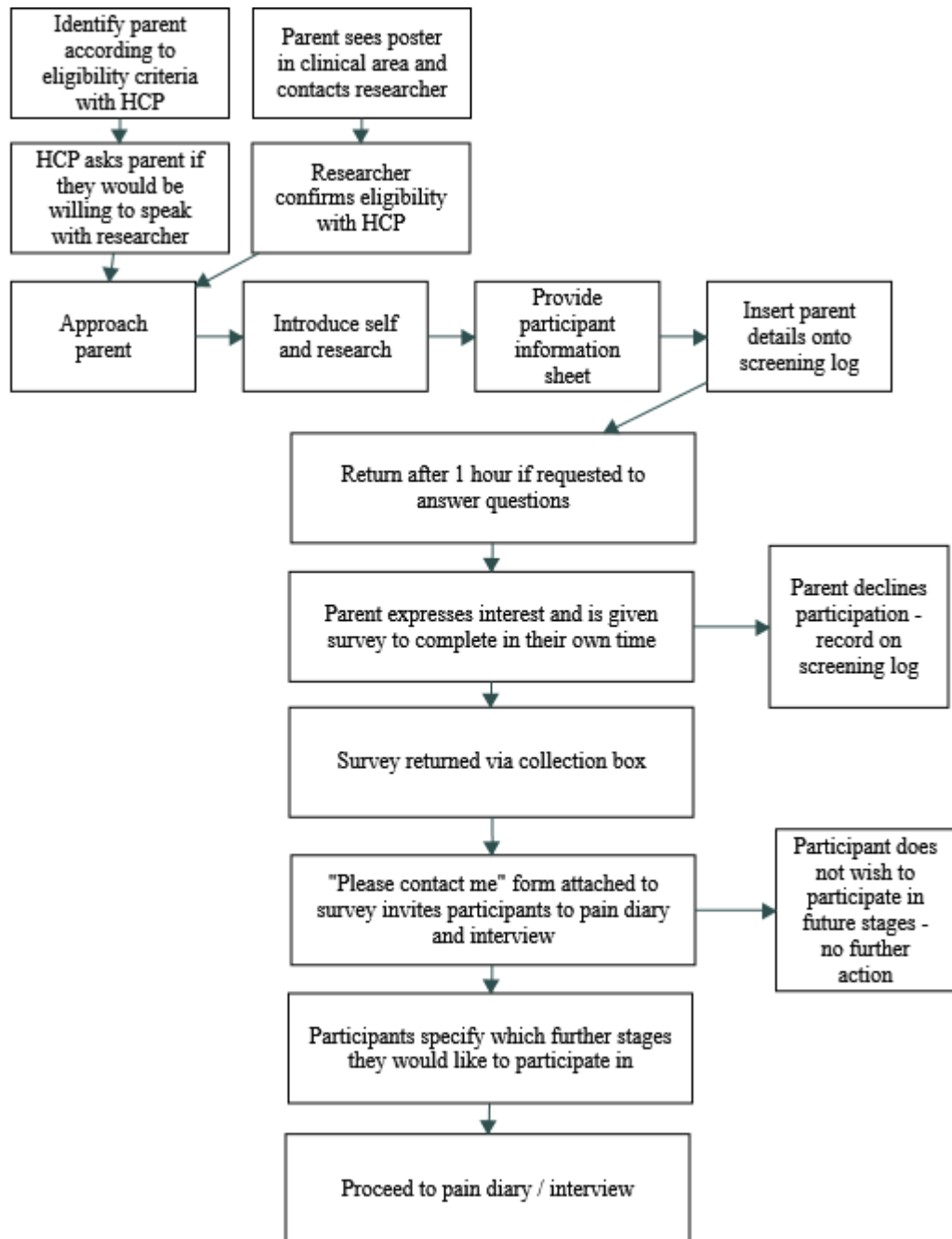


Figure 5.1: Outline of the procedure from recruitment, through to survey completion

## 5.5.4 Data analysis

### 5.5.4.1 Data cleansing

A structured process of data checking and cleansing was undertaken to minimise systemic or data entry errors. Survey data were entered into Microsoft Excel where data validation rules were applied. Once entered, data were checked against original survey documents. An analysis of acquiescent response bias (Jones and Rattray, 2010) was conducted by examining response patterns of each participant individually (Cole *et al.*, 2012; Schonla and Toepoel, 2015). Participants who did not vary their responses by more than one on the Likert scale were removed. Participant



responses were examined individually for signs of misunderstanding or misreading of reverse scored questions. Once checking was complete, data were uploaded to Statistical Package for the Social Sciences (SPSS version 21, manufacturer IBM). Box and whisker plots of scales and subscales were constructed to aid an assessment of the impact of outliers. Scores were identified as outliers if they were larger than 1.5 the interquartile range. Participants who were outliers on two or more scales had their scores and demographic background data examined for any indications of misunderstanding.

#### **5.5.4.2 Participant attrition**

Characteristics of parents who declined participation were compared to participants to identify withdrawal bias. A screening log was maintained throughout data collection where child gender, parent relationship to child, and child age were recorded. No medical records were examined in this research and consequently, it was only possible to record an estimate of the child's age in one of three categories: preschool if the child was estimated to be under four years old; primary school if estimated to be age 4-11 years; and secondary school if estimated to be age 12-16 years.

Demographic information for parents who did not participate, was ascertained by subtracting the number of participants who completed the survey from the number of parents on the screening log for each variable (child gender, parent relationship and child age). These data were used to conduct a chi-square calculation to ascertain if there was a statistically significant relationship between each demographic variable (child gender, child age, and parent gender) and the decision to participate. Where parents provided a reason for non-participation, these were collated. The researcher did not return to parents after the initial interaction unless explicitly requested by the parent so reasons for attrition could not be analysed.

#### **5.5.4.3 Demographic data analysis**

Description of the sample was provided via frequencies and percentages displayed in tables. Child characteristics included age, gender, ethnicity, diagnosis, and time since diagnosis. Parent characteristics included relationship to the child, age, ethnicity, income, and education.

#### **5.5.4.4 Descriptive analysis of survey items**

In line with previous analyses of these scales (Fortier *et al.*, 2012; Twycross *et al.*, 2015d; Zisk *et al.*, 2007b, 2010), for each item, a mean score was calculated as well as a percentage response for each response on the Likert scale. For each item, percentage agreement was calculated by grouping

“strongly agree”, “agree”, and “slightly agree”. Percentage disagreement was calculated by grouping “strongly disagree”, “disagree”, and “slightly disagree”. For each participant, a mean score was calculated for each scale and sub-scale. Where data were missing, the mean score was not calculated for the relevant sub-scale and scale.

#### **5.5.4.5 Inferential analysis of survey data**

Survey data were analysed using inferential statistics. Results were accepted as statistically significant if the probability p-value was  $\leq 0.05$ . Correlations were considered small if  $\leq .3$ , medium if  $> .3$  and  $< .5$ , and large if  $\geq .5$  (Cohen and Holiday, 1982; Penn *et al.*, 2008). Scatter plots provided visual representations of scales and sub-scales (Harris and Taylor, 2008) and the Kolmogorov-Smirnov test was used to assess distribution of data. Performing multiple statistical tests can produce significant results by chance alone so the number of tests was limited (Zaykin *et al.*, 2002). The decision of which tests to perform was made based on gaining information which was likely to provide inferences which could be applied in clinical practice. Healthcare professionals have access to child factors including diagnosis, child gender, child age, and time since diagnosis. The learning curve for caring for a child with cancer is steep initially and plateaus over time (Rodgers *et al.*, 2018) so participant responses were collated into those who had been diagnosed less than six months ago and those who had been diagnosed more than six months ago. Although HCPs are unlikely to have access to parent factors such as household income and educational background, when they meet parents they can make a rough assessment of parent age and ethnicity. Previous research has shown ethnicity to be a factor in parents’ pain knowledge and attitudes (Fortier *et al.*, 2011a; Zisk *et al.*, 2007b). The following variables were chosen to be compared to scales and sub-scales: diagnosis; child gender; child age; time since diagnosis; parent age; and parent ethnicity. Where tests involved multiple comparisons, a Bonferroni post-hoc test was used to reduce likelihood of getting significant results by chance alone. Results of the Bonferroni post-hoc tests were reported only when there was a statistically significant result in initial tests.

Categorical data with more than two groups, including diagnosis and time since diagnosis, were analysed using a one-way ANOVA if normally distributed and a Kruskal-Wallis test if not normally distributed (Myles and Gin, 2000). Ordinal data with more than two groups, including parent age, was analysed using Spearman’s rho calculation. Interval data with more than two groups, including child age, were analysed using Pearson’s correlation calculation. Data with only two groups, including pre- and post-six-months since diagnosis, which were normally distributed were analysed using Student’s T-test. Where data with only two groups were not normally distributed, Mann-Whitney U test was applied.

## 5.6 Pain diary method

As demonstrated in Table 5.1, this data collection method contributed to answering all four research questions. Previous research in this area has used interviews (Bossert *et al.*, 1996), surveys (Zhukovsky *et al.*, 2015), and pain diaries (Fortier *et al.*, 2014; Gedaly-Duff *et al.*, 2006) to gather data on children's pain at home. Both interviews and surveys are limited due to not collecting data in real-time and therefore being subject to recall bias (Jones and Rattray, 2010; McKenna *et al.*, 2010). Pain diaries are easy to complete and offer a mechanism for collecting data in real-time over a long period of time. Pain diaries are convenient for both researcher and participant, and are completed in the researchers' absence which minimises social desirability bias (Jones and Rattray, 2010). For adults with cancer, pain diaries provided high levels of accuracy when assessing pain at home due to the removal of recall bias (de Wit *et al.*, 1999). Pain diaries were valid and sensitive in recording fluctuations in pain across 4-8 cycles of chemotherapy (Geddes *et al.*, 1990). In children with cancer, pain diaries have been used to assess pain (Fortier *et al.*, 2014; Gedaly-Duff *et al.*, 2006) and to measure parents' pain management at home (Fortier *et al.*, 2014). This evidence supports use of pain diaries as an investigative tool for assessing children's cancer pain at home.

### 5.6.1 Sample

As with the survey, an ideal sampling strategy for minimising bias would be a random sample selected from a sample frame (Parahoo, 2014; Teddlie and Tashakkori, 2009), which would be unlikely to yield a sufficient sample size given study resources, time frame, and attrition. For pragmatic reasons, a convenience sample was chosen to maximise recruitment. Every parent who received a survey also received a "please contact me" form attached to the survey which they completed if they were interested in completing a pain diary. Potential participants who indicated an interest in completing a pain diary were provided with further details both verbally and via an information sheet (see Appendix 4). Consent to complete a pain diary was obtained via a consent form (see Appendix 5). Consent was re-confirmed at commencement of the pain diary. Setting, sample, and sampling technique matched the survey.

As discussed in Section 5.5.1, this research is primarily descriptive meaning that the aim of statistical analysis is to measure various parameters of interest and use these measurements to make inferences about the population. These inferences are made using the actual value and confidence interval. A sample size of 40 participants was chosen for pragmatic reasons. It was anticipated that, due to requiring a high level of involvement, fewer parents would want to participate. This sample size enabled 95% confidence intervals with +/-16% margin of error to be calculated with the exact method. A judgement was made in consultation with a statistician that 16% width was sufficiently precise to be pragmatically useful in this research.

### **5.6.2 Data collection tools**

Pain diaries were designed on Microsoft Word™ and converted into an electronic version using Bristol Online Survey (version 1, manufacturer Jisk). Face validity was obtained from academic researchers with expertise in children, pain, and cancer, as well as from experts by experience (Jones and Rattray, 2010; Parahoo, 2006; Teddlie and Tashakkori, 2009). Parents were given individual pain diaries to be completed over a one-month period. Pain diaries were provided in paper or electronic format depending on parent preference (Appendix 6). To answer question one, parents made twice-daily assessments of the location and severity of their child's pain using the NRS. They also recorded what they thought had caused their child's pain. To answer question three, free text allowed parents to describe individualised pain management techniques, their actions in response to their child's pain, and their perception of the result of their actions. If a pharmacological intervention was administered, parents were asked to record the type of pharmacological intervention and dose. If a non-pharmacological intervention was used, parents recorded this. If there was no intervention, parents were asked to state their reasons in answer to research question four. Parents were asked to record the outcome of each pain episode. Pain diaries were taken into interviews and provided reference material for discussions around how parents assess their child's pain at home. In this way, pain diaries also contributed to answering question two.

An assessment of the likely participant burden of pain diaries was made by examining completion rates in previous studies which have used pain diaries with cancer patients. Table 5.2 shows that for adults, this tool has been used for up to three months with acceptable rates of compliance. In children, this tool has been successfully administered for up to 14 days (Fortier *et al.*, 2014). Extending this duration to one month enabled patterns of pain and pain management data to be collected which aimed to give greater insight into children's cancer pain at home over time.

Table 5.2: Completion rates in studies utilising pain diaries

<b>Study</b>	<b>Population</b>	<b>Time frame</b>	<b>Compliance</b>
Van Berge Henegouwen et al. (1999)	Adults	3 months	95% of participants completed more than 95% of items
De Wit et al. (1999)	Adults	2 months	85.9% compliance
De Wit et al. (1997)	Adults	2 months	85.6% compliance
Gedaly-Duff et al. (2006)	Children	3 days	100% compliance
Fortier et al. (2014)	Children	14 days	45/55 participants completed 100%
Fortier et al. (2016)	Children	10 days	97.9% compliance

### **5.6.3 Procedure**

Figure 5.2 outlines pain diary procedures. To encourage completion, an important step in this process was meeting with participants prior to pain diary commencement to ensure they understood the study's importance and their contribution (Morren *et al.*, 2009). Parents were given choice over the mode in which they completed pain diaries and were able to see each version before deciding. Pain diaries commenced when the child was discharged from hospital. If the child was admitted to hospital during the period of pain diary data collection, parents were advised to note this but not make further recordings whilst their child was in hospital because this research aimed to investigate pain at home. Pain diary procedures were piloted by three participants recruited using the same methods as the main study. As piloting did not produce any changes in procedures, these three participants were included in the study sample.

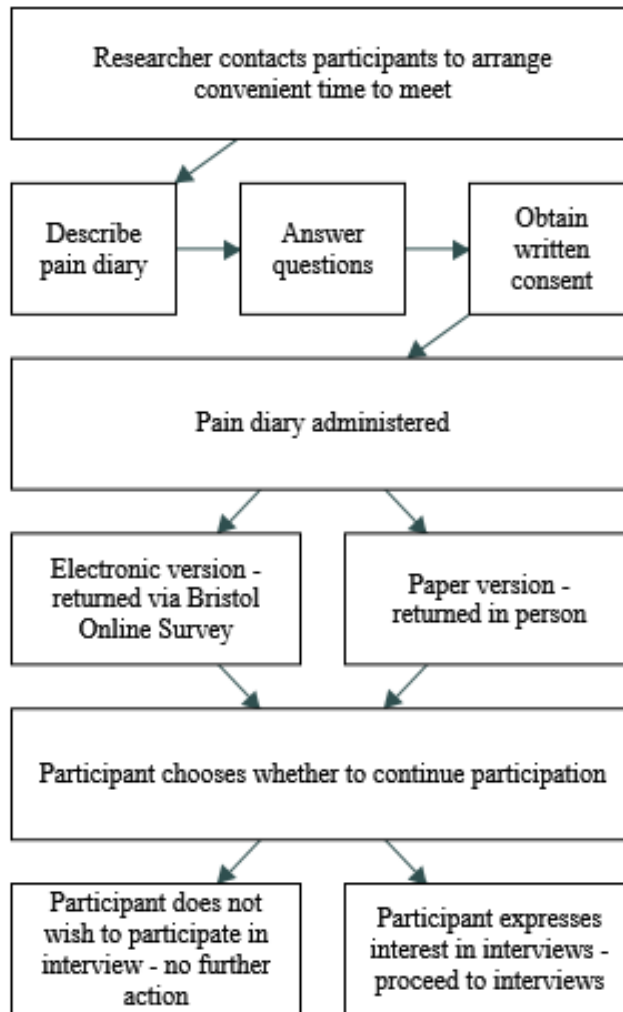


Figure 5.2: Procedure for pain diary

#### 5.6.4 Data analysis

Data entry, data checking, and choice of inferential statistics matched those used in analysis of survey data. Pain diary participants were compared to participants who completed only the survey in terms of child gender, child age (pre-school, primary or secondary school), parent gender and child diagnosis. As with survey analysis, a description of the sample was provided using descriptive statistics. Data were analysed on two levels: episode data consisted of each individual diary entry; and aggregate data consisted of summary data for each participant. For each child, number of episodes of zero pain, pain of one or more, and clinically significant pain (pain score of more than three on the NRS) (Fortier *et al.*, 2014), were calculated. Maximum pain and mean pain were calculated for each child and compared to diagnosis and time since diagnosis.

Choice of inferential statistics was based on type and distribution of data as detailed in Section 5.5.4.5. Parent responses to pain were grouped into pharmacological interventions, non-pharmacological interventions, a combination of pharmacological and non-pharmacological interventions, and no action. Pharmacological interventions were further grouped into analgesic drugs, topical, antiemetic, laxative, and antidiarrheal. Non-pharmacological interventions and pain cause were grouped into nominal data. Frequency of episode and mode per child of pain location, cause of pain, parent response, pharmacological interventions administered, analgesic drugs administered, and non-pharmacological interventions were calculated. Number of pharmacological interventions, analgesic drugs, paracetamol, and morphine administrations were calculated per child. Frequencies and modes were displayed in tables, if children had more than one mode, all modes were included. Analgesic drug administrations were calculated and displayed figuratively for episodes in which children had a pain score of zero, one or more, and three or more.

Parents' responses to pain were analysed in relation to pain score. Using Kruskal-Wallis would not have been appropriate as each parent provided data for multiple episodes, so episodes cannot be considered independent (Schmider *et al.*, 2010). A univariate ANOVA was chosen because it takes into account individual parent preferences for each response without the requirement for matched data (Schmider *et al.*, 2010). Although it assumes normality, this test is robust even with departure from normality. Using a quantitising technique in which qualitative data are converted to numerical data (Bazeley, 2009; O'Cathain *et al.*, 2010), outcome of each episode as recorded by parents were converted into ordinal data by categorising according to improvement or no improvement in pain. Parent responses were calculated and displayed figuratively for episodes which were recorded as improvement and no improvement. Pharmacological intervention dosages were calculated, and results ranked into ordinal data depending on whether parents had administered a lower, optimum, or higher dose than that which is advised in the British National Formulary (Paediatric Formulary Committee, 2016).

#### **5.6.4.1 Qualitative analysis**

Free text responses to "If no action is taken or required, please say why" and "any other comments", provided participants with an opportunity to expand on responses (Jones and Rattray, 2010). These were analysed using content analysis with low abstraction degree and low interpretation level (Graneheim *et al.*, 2017). An alternative analysis approach is thematic analysis (Braun and Clarke, 2006). Due to the brevity of responses and lack of rich qualitative data, thematic analysis would not have been appropriate. Similarly, framework analysis was a potential alternative method of analysis which would not have been appropriate as the size of the dataset was insufficient to identify a robust framework and neither mapping nor interpretation would have been

possible (Ward *et al.*, 2013). Content analysis was selected to give appropriate attention to participants' responses, whilst ensuring data were not unduly weighted or overemphasised. Responses were uploaded to NVivo™ (Version 10, QSR International), read through entirely to enable understanding of the data as a whole, coded, and then grouped into categories and subcategories. Responses were not coded if they had contributed to a data field which had already been analysed, for example, if the comment addressed the cause of pain.

## **5.7 Interview method**

Qualitative data collection using interviews aimed to understand the qualitative aspects of all four research questions (Table 5.1) with a focus on subjective norms and PBC (Section 4.3). According to TPB, subjective norms have an influence on intention and consequential behaviour. Subjective norms are a combination of an individual's motivation to comply with significant others and their normative beliefs. Normative beliefs are a product of what an individual believes significant others in their life think they should do (Ajzen, 1991). Perceived behavioural control is an individual's perception of their ability to perform a behaviour stemming from beliefs about barriers and ease with which a behaviour is likely to be performed (Ajzen, 2005, 2011). Quantitative tools exist to measure these concepts in other areas (Ham *et al.*, 2015; Walsh *et al.*, 2009) but no tools exist for measuring these concepts for parents managing their child's pain at home. A key feature of TPB is that concepts within it are situation specific (Ajzen, 2011) so using alternative tools would not have been appropriate. An additional consideration is that quantitative methods are not well suited to subject areas requiring elaboration, causal explanations, or increased understanding of barriers and facilitators of phenomena (Parahoo, 2006).

One alternative could be to conduct focus groups with this population (Krueger, 2008). Whilst this method could eliminate many pitfalls described above, there are two disadvantages for this research. Firstly, parents of children with cancer hold many additional caring responsibilities making their availability limited (Molinaro and Fletcher, 2018). It is important to use methods of data collection which offer flexibility to participants to avoid restricting recruitment and impeding patients' and participants' lives (Todd, 2010). Secondly, the shared nature of focus groups meant that it would not be possible to discuss participant responses from pain diaries in these groups without breaking participant confidentiality (Parahoo, 2006). Therefore, qualitative research interviews were chosen to investigate subjective norms and PBC. Interviews allowed participants to select time and location of data collection for their own convenience thereby providing a pragmatic solution to maximise recruitment (Todd, 2010). Interviews were private and tailored to each participant according to their responses from pain diary without breaking participant confidentiality (Parahoo, 2006). Interviews collected in-depth information and were appropriate for



exploring phenomena about which there is little information (Krueger, 2008; McKenna *et al.*, 2010).

### 5.7.1 Sample

Interview participants were recruited from the pain diary sample. If parents wished to participate in an interview, they completed a “Please contact me” form which was attached to the survey and pain diary. Information sheets provided to potential participants who expressed interest in interviews by completing the form prior to pain diary participation also contained information on interviews (Appendix 4). This information was reiterated prior to obtaining interview consent which was confirmed by completion of a consent form (Appendix 6). Consent was re-confirmed verbally prior to starting each interview. Parents who expressed interest in interviews but who did not get to participate due to the purposive sampling method adopted were contacted individually, thanked for their time and interest, and offered an opportunity to receive a copy of results at the end of the study.

Sampling in qualitative research prioritises data which the sample is likely to yield over sample size or representativeness (Mason, 2010). Ideally, sampling should cease only when no new themes emerge, a phenomena known as data saturation (Smith and Noble, 2014). The sampling strategy was chosen to make the most of limited resources by obtaining the greatest depth and breadth of information from few participants (Teddlie and Tashakkori, 2009). Purposive sampling was used to maximise resources and obtain a representative sample. Table 5.3 displays the sampling strategy created to select participants based on their likely contribution (Heavey, 2014) according to the research question (Parahoo, 2006). Selection maximised variation and gained a broad understanding of phenomena (Teddlie and Tashakkori, 2009).

Table 5.3: Intended sampling frame

	<b>Birth – 3 years</b>	<b>4-7 years</b>	<b>8-16 years</b>	<b>Total</b>
First six months	3	3	3	9
After six months	3	3	3	9
Total	6	6	6	18

Participants were selected according to stage of treatment and age of child. Selection according to stage of treatment was important because evidence suggests family informational needs evolve throughout the cancer treatment trajectory (Woodgate and Degner, 2003). It was likely that parents’

pain management abilities, barriers, and facilitators, would also change throughout the treatment trajectory. Age selection was based on previous literature investigating children's cancer pain at home (Bossert *et al.*, 1996; Fortier *et al.*, 2014; Gedaly-Duff *et al.*, 2006; Zhukovsky *et al.*, 2015) as well as Piaget's stages of cognitive development (Piaget *et al.*, 1952). Key literature quantifying parents' management of children's cancer pain at home has not represented children under the age of four, so this group was targeted in the sampling strategy in an attempt to bridge gaps in research. According to Piaget's stages of cognitive development (Piaget *et al.*, 1952), children's cognitive development transitions from the pre-operational stage to concrete operational stage around the age of seven. On entering the concrete operational stage, children are able to make generalisations and use logic and inductive reasoning. Before this stage of cognitive development, children will experience, process, express, and communicate pain differently to children who have transitioned through this stage (Gaffney and Dunne, 1986, 1987; Twycross, 1998). It was important that parents of children in both stages were represented in interviews.

### **5.7.2 Data collection tools**

Due to the paucity of knowledge on this topic, a highly structured interview was not considered appropriate (Parahoo, 2014; Todd, 2010). Interviews prioritised exploration above consistency leading to a semi-structured (McKenna *et al.*, 2010), focused-qualitative (Parahoo, 2006), or lightly structured schedule (Wengraf, 2001). The interview schedule (Appendix 7), was derived from background literature, experts by experience, and guided by TPB (Ajzen, 1991). It was designed to enable an understanding of environmental and social factors which act as barriers to behaviour change (NICE, 2007).

Each participants' completed pain diary was taken to the interview to prompt responses and allow the participant to clarify and elaborate on their actions as recorded. At the end of the interview, the researcher summarised the discussion and participants were given opportunity to provide feedback as well as to clarify and correct the summary (Thomas, 2006). The interview schedule received face validity from several experienced researchers with expertise in cancer, pain, and paediatrics as well as experts by experience (Jones and Rattray, 2010; Parahoo, 2006; Teddlie and Tashakkori, 2009).

### **5.7.3 Procedure**

Figure 5.3 details interview procedures. A process consent model was adopted (Dewing, 2007) with consent obtained at point of recruitment as well as prior to the interview (Appendix 8). The

researcher contacted participants at least one day prior to the interview to ensure their continued availability. Interviews were conducted in a location of the participants' choosing which was accessible, comfortable, and minimised risk of disruption (McKenna *et al.*, 2010). Wherever possible, interviews were conducted face-to-face to allow observation of body language and more appropriate response to expressions of emotion (McKenna *et al.*, 2010). Where necessary for participants' convenience or preference, telephone interviews were undertaken. Some interviews were conducted in more than one sitting due to interruptions. This resulted in one interview being conducted partly in hospital and partly in the participants' home. Interviews were audio recorded using a digital recorder. The interview schedule was tested in a pilot with the same three participants who piloted the survey. Procedures followed the main study and pilot participants were given an opportunity to provide feedback. This feedback may have resulted in changes of protocol prior to study commencement, but as no changes were made, pilot data were included in results.

Interviews commenced with the researcher reminding participants that there were no right or wrong answers, ensuring they understood their right to withdraw (McKenna *et al.*, 2010), and explaining the purpose of note-taking (Parahoo, 2006). Initial opening questions allowed the participant and researcher to build rapport, gave participants freedom to tell their story, and provided the researcher with context for the remainder of the interview (Todd, 2010). Questions then addressed less sensitive issues relating to behaviour, experience and knowledge. Towards the end of the interview, questions were asked addressing more sensitive topics such as opinion, beliefs, and feelings (McKenna *et al.*, 2010). Research question one was answered by asking how pain had affected the child with cancer. Research question two was answered by asking probing questions to clarify how parents knew their child was in pain. Research question three was answered by asking about helpful techniques used to manage pain. Research question four was answered by asking about barriers and facilitators to pain management. Prompts were used to clarify and test assumptions and keep discussions on track (McKenna *et al.*, 2010). Interviews explored participants' PBC and subjective norms (Ajzen, 1985). Subjective norms were addressed by asking participants about the individuals who supported them in managing their child's pain at home. This question was left open-ended to allow participants to describe those closest to them as well as any influence of wider culture. Perceived behavioural control is a measure of the resources and opportunities which relate to a behaviour and may reflect realistic barriers to behaviour (Ajzen, 2005). Ajzen discusses several internal and external factors which may contribute to PBC (Ajzen, 1985). Internal factors were addressed with one question. External factors were addressed with two questions.

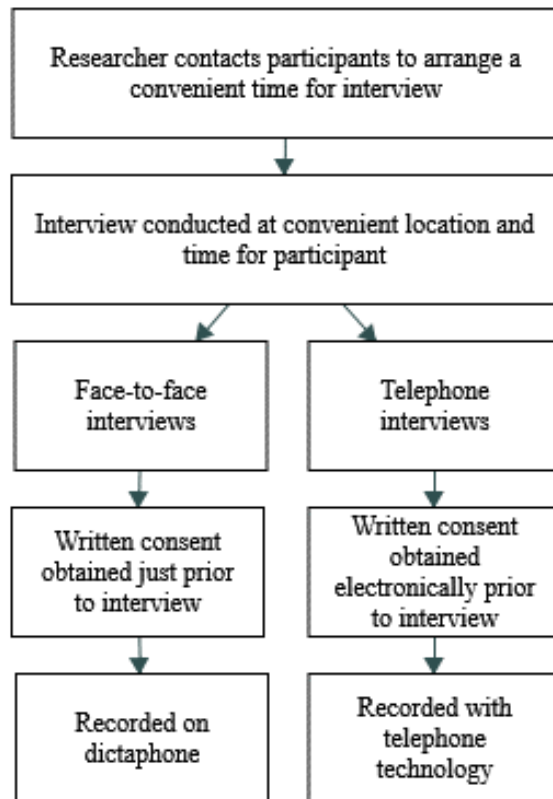


Figure 5.3: Procedure for conducting interviews

#### 5.7.4 Data analysis

Several types of qualitative analysis were considered. Content analysis may have led to a loss of complexity and meaning stemming from data, and would not have provided the depth or holistic interpretation that this analysis aimed to achieve (Schreier, 2014; Silverman, 2010). Framework analysis may have been appropriate but was not preferred as it has been criticised for tempting researchers to take shortcuts (Ward *et al.*, 2013). This analysis aimed to stay as close to data as possible by using an iterative approach of constant return to data which may have been hindered by framework analysis. An inductive, iterative approach was chosen for this analysis as it is a relatively under-researched area with no clear theory from which to derive a framework for deduction. Due to its flexibility and applicability across a range of theoretical frameworks, thematic analysis was considered an appropriate analytic approach.

Qualitative data were analysed using NVivo™ (Version 10, QSR International) following the phases outlined in Table 5.4 (Braun and Clarke, 2006). Data were transcribed verbatim by the researcher which increased opportunity for reflexivity (Jootun *et al.*, 2009; McKenna *et al.*, 2010). Transcriptions were checked against original recordings to ensure accuracy (Braun and Clarke, 2006; Parahoo, 2006). A summary of each transcript was written. Demographic data were used to

contextualise transcripts. Thematic analysis involves searching data for repeated patterns and understanding how these patterns fit within a dataset to tell a story (Howitt and Cramer, 2010). Researchers have advocated drawing a map or diagram to represent themes pictorially (Clarke *et al.*, 2015; Willig, 2013). This technique was used to help understand relationship between themes, make sense of data, and move the analysis beyond simply summarising data. Attention was given to ensuring data were represented as a whole whilst answering each research question. In addition, analysis involved searching for evidence of attitudes, subjective norms, and PBC (Ajzen, 2011).

Table 5.4: Phases of Thematic Analysis

Phase	Action
Familiarisation	Transcribing, reading, re-reading, noting initial ideas
Generating initial codes	Coding the entire dataset systematically, collating each code
Searching for themes	Collating codes into potential themes
Reviewing themes	Relating coded extracts to the dataset as a whole
Defining and naming themes	Ongoing refinement, written definitions of themes
Producing the report	Relating to the research question and literature

## 5.8 Data integration method

Successful mixed methods data integration is a result of good quality analysis of individual datasets as well as good quality data integration (Creswell and Plano Clark, 2011; Teddlie and Tashakkori, 2009). Sections 5.5, 5.6, and 5.7 detail how this research has been designed to achieve good quality data for each method of data collection. Mixed methods data integration was a separate stage of analysis to quantitative or qualitative analysis.

This section discusses integration at design level, method level, and interpretation/reporting level (Fetters *et al.*, 2013). Figure 5.4 displays pictorially the process from each data collection method through analysis to inference and meta-inference generation. At the design level, integration was achieved through use of a convergent, parallel design (Creswell and Plano Clark, 2011). At the method level, data integration was achieved through “merging”, an approach which is most common for convergent parallel designs (Fetters *et al.*, 2013). Within this method, datasets were brought together for analysis and comparison after separate quantitative and qualitative analysis. Completed pain diaries for each interview participant contributed towards data collection during interviews which constituted within-participant integration (Fetters *et al.*, 2013). This approach should not be confused with “connecting” where the sample for one data collection method is obtained based on results of a previous data collection method (Fetters *et al.*, 2013). In this

research, sample selection was based only on completion of previous data collection methods, not on results or performance of previous data collection methods. Neither should the approach be considered “building” in which one database informs data collection for another because an individual pain diary does not constitute a database (Fetters *et al.*, 2013). At interpretation and reporting level, matrices, joint display, and narrative using the contiguous approach were used in analysing and presenting results (Creswell and Plano Clark, 2011; Fetters *et al.*, 2013; Guetterman *et al.*, 2015).



Figure 5.4: Analysis process and meta-inference generation

### 5.8.1 Statistical integration of survey and pain diary data

Survey and pain diary responses were compared for participants who completed both. Distribution of scales and sub-scales were analysed using the Shapiro-Wilk test which is appropriate for

samples less than 50 (Razali and Wah, 2011). Statistical tests chosen matched those used in survey and pain diary analysis. Maximum and mean pain scores, and number of episodes of clinically significant pain were statistically compared to scales and sub-scales. For each child, the number of episodes in which parents administered pharmacological interventions, non-pharmacological interventions, a combination of pharmacological and non-pharmacological interventions, or took no action, were compared to scales and sub-scales. The total number of analgesic drugs, paracetamol, and morphine doses administered were compared to scales and sub-scales for each child.

### ***5.8.2 Meta-inference generation through integration of datasets***

Survey, pain diary and interview data integration was conducted in three phases. Initially a matrix structured around each data collection method facilitated integration of findings from three large datasets (Dickson *et al.*, 2011; Miles and Huberman, 1994). Each inference for which there was evidence in more than one dataset was represented in a row. Inferences relating to each method of data collection were listed in the first column. For each inference, convergences and divergences from other datasets were added in subsequent columns by returning to each dataset. In the final column, meta-inferences stemming from matrices were listed (Bazeley, 2009; Dickson *et al.*, 2011; Miles and Huberman, 1994). This initial phase served to assist the researcher in organising a vast amount of data and helped identify important findings for discussion

The second phase of integration involved joint display of findings structured around research questions (Creswell and Plano Clark, 2011; Fetters *et al.*, 2013; Guetterman *et al.*, 2015). A single table was constructed which provided findings from each data collection method in relation to each research question (Creswell and Plano Clark, 2011). For each research question, convergent and divergent results were presented alongside potential explanations for findings. This second phase enabled generation of meta-inferences and provided the basis for the third phase.

Potential explanations from the joint display were used for the third phase of integration which occurred in Chapter 8. Meta-inferences stemming from convergences and divergences were discussed in light of literature and theory was built around these inferences (Teddlie and Tashakkori, 2009). Integration through narrative was achieved through a contiguous approach in which findings from each data collection method were brought together in a single report with different sections for each research question (Fetters *et al.*, 2013). Different data collection methods examined different aspects of the same phenomena so to weave or thread themes between them would be incongruous with the design. Throughout integration, divergences were actively pursued as seeking reasons for divergences could enhance theory (Teddlie and Tashakkori, 2009).

Differences in the source of each inference provided context for inferences and aided development of meta-inferences which could explain both divergences and convergences. This process eventually led to theory development.

## **5.9 Strategies for minimising threats to validity**

Mixed methods researchers recognise the need to evaluate inferences derived from quantitative data using quantitative frameworks, as well as evaluating inferences derived from qualitative data using qualitative frameworks, and to identify the extent to which meta-inferences are credible (Teddlie and Tashakkori, 2009). This section of the thesis outlines how this research has been designed to accomplish each of these three assessments.

### **5.9.1 *Validity in quantitative data collection and analysis***

Teddlie and Tashakkori (2009) describe four types of validity in quantitative research: statistical conclusion validity, internal validity, construct validity, and external validity. In this section, each type of validity is described along with ways in which this research has been designed to increase validity. Statistical conclusion validity was enhanced by: minimising the number of statistical tests performed to limit the likelihood of significant results by chance alone (Zaykin *et al.*, 2002); including a Bonferroni post-hoc test where multiple tests were required; ensuring assumptions of statistical tests were not violated (Teddlie and Tashakkori, 2009); and ensuring the study was not left underpowered by maintaining sample sizes sufficient to produce pragmatically useful confidence intervals. Internal validity was enhanced by: pilot testing procedures to ensure their acceptability to participants (Sections 5.5.3, and 5.6.3); monitoring attrition and characteristics of participants who withdrew with chi-squared analyses; and using previously validated tools with good internal consistency (Chambers *et al.*, 1997; Forward *et al.*, 1996; Zisk *et al.*, 2007a, 2010). Similarly, using tools which have previously demonstrated good construct validity increases construct validity in this research (Forward *et al.*, 1996; Zisk *et al.*, 2007a). Finally, external validity was enhanced by using broad inclusion criteria (Polit and Beck, 2010). Including all children with cancer regardless of pain manifestation meant findings could be applied to a general childhood cancer cohort. In addition, collection of several demographic characteristics meant a rich description of sample could be provided to allow an assessment of external validity.

### **5.9.2 *Rigour in qualitative data collection and analysis***

This section outlines procedures selected to increase rigour and provides rationale for each. Although debates continue, there is no set criteria for ensuring rigour in qualitative research (Noble and Smith, 2015; Rolfe, 2006). Despite contradictory messages, researchers must take



responsibility for ensuring rigour (Rolfe, 2006). Three principles were used in selection of processes to increase rigour: transparency in decision making; strategies during data collection; and multiplicity of strategies. Firstly, the principle of transparency (Kuper *et al.*, 2008; Morse *et al.*, 2002; Rolfe, 2006) was used in three ways. Transparency of decision making as described in this section of the thesis, where justification for each strategy selected is clearly described.

Transparency was increased through record keeping and providing an audit trail of decision making processes (Kuper *et al.*, 2008; Noble and Smith, 2015; Slevin and Sines, 2000; Smith and Noble, 2014). Use of quotes which were representative of participants views, spreading out quotes between participants, and choosing most typical rather than most interesting quotes increased transparency further (Holloway and Wheeler, 2013).

Secondly, strategies which increase rigour were implemented during data collection as opposed to only post-hoc verification (Morse *et al.*, 2002). This included reflexivity and participant feedback. Qualitative data are resultant from interactions between participant and researcher. Researchers alter data collection by what they bring to interactions. Reflexivity is the practise of identifying how a researchers' actions, beliefs, biases, and presence impact data (Berger, 2013; Parahoo, 2006). Reflexivity accounts for researcher bias and transforms subjectivity into a positive tool (Berger, 2013). In this research, reflexivity was exercised by frequent supervision, keeping a journal of self-reflection (Berger, 2013; Jootun *et al.*, 2009), reflecting on actions, and considering personal prejudices and assumptions throughout data collection (McKenna *et al.*, 2010). Participant feedback was conducted as part of the interview schedule to further increase rigour (Parahoo, 2006).

The final principle is that of multiplicity, meaning that several strategies were used to enhance rigour and its assessment (Morse *et al.*, 2002). These multiple strategies included the refutability principle, thick description, and peer checking. The refutability principle involves seeking to disprove each inference before confirming its objectivity (Silverman, 2009). This was achieved via comprehensive data treatment where the full dataset was analysed, checking for internal coherence, internal consistency, and distinctiveness of themes (Braunack-Mayer, 2002). Searching for deviant cases which contradict inferences, further enhanced the refutability principle (Noble and Smith, 2015; Silverman, 2009). Once a coherent model was produced, each case was returned to and examined in light of the model. Deviations from the model were used to alter it until these procedures led to a set of rules which could explain the dataset as a whole and increase theoretical understanding (Kuper *et al.*, 2008). Thick description of the sample enabled transferability of judgements and the most typical rather than most interesting quotes from participants were chosen in reporting (Kuper *et al.*, 2008; Slevin and Sines, 2000). Peer checking involved having a second

researcher (in this case a doctoral supervisor) code the first four interviews independently and results of both student and supervisor coding compared (Creswell and Plano Clark, 2011). Rigour was further enhanced in supervision sessions during which codes were challenged and evidence for codes and model were questioned.

### ***5.9.3 Validity in mixed methods research***

The term “validity” is used in reference to mixed methods research due to its acceptability to both qualitative and quantitative researchers (Creswell and Plano Clark, 2011). Potential threats to the validity of mixed methods research as well as strategies to minimise threats differ depending on study design. Table 5.5 displays potential threats to validity in data collection, data analysis, and interpretation as well as strategies recommended for minimising the threat and specific strategies used in this research.

## **5.10 Ethical considerations**

### ***5.10.1 Ethical approval***

Ethical approval was granted from the Health Research Authority (16/NS/0121, Appendix 9) following governance and approval from The Royal Marsden NHS Foundation Trust (CCR4569/AM1702/04, Appendix 10). As per university procedures, this study was also ratified by London South Bank University Research Ethics Committee (UEP1116, Appendix 11).

### ***5.10.2 Informed consent***

The researcher completed both online and face-to-face informed consent training courses. Written informed consent was obtained from participants for pain diary and interview (Silverman, 2010). A combination of both verbal and written information was provided (Nishimura *et al.*, 2013) which was carefully prepared to ensure it enabled participants to make a truly informed decision (Silverman, 2009). Written information was clear and concise with a level of detail which provided important information but was not overwhelming. Verbal information followed similar principles with the researcher being friendly and approachable without coercing or putting pressure on participants (Health Research Authority, 2018). Every effort was made to ensure participants were aware of their right to withdraw (Silverman, 2010). Information provided to participants was overt and honest as this is ethically preferable and there was no justification for covertness (Silverman, 2010; Teddlie and Tashakkori, 2009).

Table 5.5: Potential threats to validity adapted from Creswell and Plano Clark (2011)

<b>Potential validity threats when merging data</b>	<b>Strategies for minimising the threat</b>	<b>Specific strategies utilised in this research</b>
<i>Data collection issues</i>		
Selecting inappropriate individuals for qualitative and quantitative data collection.	Drawing quantitative and qualitative samples from the same population increases data comparability.	Parallel mixed methods sampling (Teddlie and Tashakkori, 2009), sampling for convergent designs (Creswell and Plano Clark, 2011), mixed methods sampling strategy (Teddlie and Yu, 2007) used.
Introducing potential bias into one data collection through the other data collection.	Use separate data collection procedures and collect data at the end of an experiment.	Ordering of data collection procedures: survey and pain diary before interview so qualitative data collection could not affect quantitative data collected.
Collecting two types of data that do not address the same topics.	Address the same question (parallel) in both quantitative and qualitative data collection.	Qualitative and quantitative data collection did not claim to measure the exact same phenomena, rather different aspects of the same phenomena were measured by different data collection procedures. Transparency of the purpose of each data collection method minimised threats to validity.
<i>Data analysis issues</i>		
Using inadequate approaches to converge the data.	Develop a joint display with categorical data and qualitative themes or use other display configurations.	Meta-inference generation occurred through use of matrices and joint display.
Making illogical comparisons of the two results of analysis.	Supporting statistical results with qualitative quotes.	Although seeking quotes to match statistical results could have been conducted effectively, the researcher sought to provide most typical quotes and examined divergences as well as convergences.
<i>Interpretation issues</i>		
Not resolving divergent findings.	Using strategies such as gathering more data, reanalysing the current data, and evaluating the procedures.	Divergent findings revealed deeper inferences and were resolved by examining potential explanations.
Not discussing the mixed methods research questions.	Address each mixed methods question.	Each question is discussed with reference to results, literature and theoretical framework in the discussion chapter.
Giving more weight to one form of data than the other.	Use procedures to present both sets of results in an equal way or provide a rationale for why one form of data provided a better understanding of the problem.	Data are presented in matrices, joint display and narrative. Each data collection method provides information on a different aspect of each research question. Rationale is provided for how each data collection method contributes to each question.

Parents were given time between receiving information and commencement of participation to allow them to consider their participation, talk with friends and family, and identify areas which required clarification (Todd, 2010). The Health Research Authority state that duration between approach and consent should be based on participant burden (Health Research Authority, 2018). With one exception, this time frame was a minimum of 24 hours. The single exception was survey administration. Parents were approached and given information in an outpatient clinic at The Royal Marsden NHS Foundation Trust. Initial approach and provision of information was followed by the researcher returning, on parents' request, approximately one hour later to provide an additional opportunity to ask questions and raise concerns. During this time, the researcher assessed the extent to which parents understood the study. Returning one hour later to provide parents with a survey whilst they were present in hospital negated the need for them to return to hospital at a later date to collect a survey. Provision of the survey did not mean the parent needed to participate and afforded them the right to choose their preferred time and location of participation. This is in line with guidance issued by the National Research Ethics Service (NRES, 2016). Consent for the survey was implied by completion, a model which has been shown to reduce inequalities in recruitment of survey research (Unger *et al.*, 2004).

### ***5.10.3 Anonymity and confidentiality***

Procedures were designed to promote participant anonymity and confidentiality. Identifiable information was not collected in surveys unless participants wished to continue to further data collection methods. In this case, it was necessary to collect identifiable information to enable responses of individuals to be linked between data collection methods. In this case, although anonymity could not be maintained, confidentiality was maintained. Mechanism of return facilitated participant anonymity.

Confidentiality was ensured by storing information securely in areas accessible only via a swipe card. Electronic data were stored on password protected computers. Completed questionnaires, consent forms, and paper pain diaries were stored in a locked filing cabinet. Only the researcher had access to identifiable data (Green and Thorogood, 2013; Silverman, 2010). Participants were assigned a unique ID or pseudonym to aid analysis whilst maintaining confidentiality. Identifiable information was removed from transcripts. Participants may be able to identify themselves in quotes used in publications, but identifiable information was removed, so others will not be able to identify participants. Quantitative information was collated so participants will not be able to identify themselves in quantitative aspects of publications.

#### ***5.10.4 Ethical conduct of qualitative research interviews***

Interview technique was enhanced by practice, prior experience, and pilot interviews. Whilst the purpose is not therapeutic (Parahoo, 2006), there is growing recognition of therapeutic benefits experienced by participants during research interviews (Coombs *et al.*, 2016; Opsal *et al.*, 2016; Rossetto, 2014). Research interviews can mirror therapeutic interviews (Birch and Miller, 2000; Silverman, 2010). Being cognisant of this, the researcher used strategies to maximise benefit and minimise harm (Fine, 2003). Attention was given to building rapport through humour, attention and interest, being responsive to mood and mindful of posture (McKenna *et al.*, 2010). Care was taken to ensure rapport was not used in a manipulative way to encourage unwitting disclosure (Mitchell and Irvine, 2008; Parahoo, 2006; Silverman, 2010). A flexible approach was taken to personal sharing or disclosures and attention was given to language used, to ensure a non-judgemental approach (McKenna *et al.*, 2010). Interviews were conducted with awareness that the researcher was not part of the study population (Berger, 2013).

#### ***5.10.5 Participant and researcher safety during interviews***

Participant and researcher safety are paramount. All participants were vulnerable due to the burden of having a child with cancer and the topic sensitive due to parent distress associated with seeing their child in pain (Forgeron *et al.*, 2006; Pöder *et al.*, 2010). Techniques to reduce emotional distress included being led by participants, allowing silence, moving on from distressing subjects, and creating breaks (Mitchell and Irvine, 2008; Silverman, 2010). The researcher was aware of local procedures to access support for participants including psychologists and social workers. Participants were made aware of this prior to interview commencement.

Due to being a lone worker, attention was given to researcher safety. The researcher was familiar with and adhered to The Royal Marsden NHS Foundation Trust policy for lone workers. A process was set up which minimised risks to the researcher due to working alone. When conducting interviews outside the clinical environment, the researcher ensured a colleague knew the date, time, and location of each interview. The researcher telephoned the colleague on arrival, within two hours of arrival, on departure, and when arriving home. If the colleague did not receive a phone call at an expected time, they would call the researcher. Each phone call confirmed that the researcher was safe.

### **5.10.6 Nurse as researcher**

As a registered children's nurse, the researcher adhered to the Nursing and Midwifery Council Code at all times (NMC, 2015). The researcher was cognisant that the nursing role could come into conflict with the researcher role (Holloway and Galvin, 2016). During interactions with participants, the researcher exercised transparency about her dual roles, capacity, and limitations of each. Both roles were acknowledged, and the purpose of interactions made clear. If conflict arose, this was discussed on a case-by-case basis with supervisors who were also registered children's and adult nurses (McKenna *et al.*, 2010). Debriefing sessions with supervisors allowed the researcher to deal with emotions, increase reflexivity, and maintain researcher wellbeing (Boden *et al.*, 2015; Jootun *et al.*, 2009; Mitchell and Irvine, 2008; Parahoo, 2006).

The dual role of nurse and researcher may have caused problems for participants (Holloway and Galvin, 2016). Parents may have known the researcher in a nursing capacity and consequently felt obliged to participate. Three strategies were implemented to minimise this possibility. Firstly, information provided to participants emphasised the voluntary nature of participation and that participation would not affect their child's care. Secondly, after initial approach, participants were the sole instigator of participation: information was left with participants and the researcher did not re-instigate contact unless requested to do so by participants. Finally, evidence suggests participants use a variety of indirect strategies to opt out of participation if they do not feel able to do so directly (Mitchell and Irvine, 2008). The researcher was aware of strategies such as not responding to phone calls or cancelling appointments and responded sensitively. Knowledge that the researcher was a nurse may have meant participants asked questions outside the scope of the study (Holloway and Wheeler, 2013). Reasonable effort was made to anticipate potential questions and provide resources for participants where available and appropriate. Participants were shown respect and questions outside the scope of the study were not ignored. Participants were provided information regarding support such as counselling services where appropriate (Birch and Miller, 2000; Mitchell and Irvine, 2008).

## **5.11 Summary**

This chapter provided details of methods employed in this research, practical steps undertaken in data collection and analysis, and rationale for methodological decisions. The Theory of Planned Behaviour was utilised alongside a convergent, parallel, mixed methods design. Parents' attitudes were investigated using a survey constructed from two previously validated questionnaires. Survey methodology allowed a large number of participants to be sampled with relative convenience to participant and researcher. Participants who wished to continue participation completed a pain diary, interview, or both. Pain diaries provided a real-time measure of pain experience. Interviews

were used to investigate subjective norms and PBC. This allowed data to be gathered on a subject about which little is known. Data for each stage were analysed separately and then integrated. Ethical issues were discussed. The next chapter presents the results of these investigations.

## Chapter 6. Results

### 6.1 Overview of results chapter

This chapter presents the results of analysis of survey, pain diary, and interview data. Both survey and pain diary data are presented in a similar pattern with dataset and sample population described first followed by descriptive and then inferential statistics. Interview findings commence with a rich description of the sample. Following this, interview findings are presented in a model format in which enables understanding of relationships between themes and how they live within the dataset.

### 6.2 Survey findings

Survey results are described below. Initially data cleansing results are presented followed by results of participant attrition analysis. Descriptive statistics provide a summary of sample characteristics and report of responses to each item of the MAQ and PPEP. Finally, inferential statistics are presented.

#### 6.2.1 *Data quality and distribution*

Data cleansing processes followed procedures outlined in Section 5.5.4.1 with the purpose of minimising systemic or data entry errors. No participants were identified as having acquiescent response bias (Jones and Rattray, 2010). Neither were any participants identified as having misunderstood or misread reverse scored items. Both MAQ and PPEP scales were normally distributed, but only two sub-scales (PPEP attention seeking and MAQ avoidance) were normally distributed. As not all scales were normally distributed, tests involving both scales and sub-scales were analysed using non-parametric tests to ensure consistency and that no statistical assumptions were violated.

#### 6.2.2 *Participant response rate and assessment of response bias*

Of the 161 parents invited to participate, 101 (62.7%) returned the survey. Although recruitment stopped once 100 surveys had been received, one survey was received after this point so was included in analysis. Table 6.1 displays a chi-square analysis which was conducted to ascertain whether the method of data collection had skewed the sample demographically. Child gender, child age, and parent relationship are presented depending on whether parents chose to participate or not. When both parents were approached (n=34), this was noted on the screening log as “both”. When completing the survey, one set of parents did so together but were excluded from the chi-square



analysis because this would have resulted in cell sizes too small to analyse. No statistically significant difference in participation rate was found by gender of child, age of child, or relationship of parent.

Table 6.1: Screening demographics and chi-square calculation

	<b>Participation</b>	<b>Non-participation</b>	<b>Total screened</b>	<b>Chi-square</b>	<b>df</b>	<b>p-value</b>
<b>Child gender</b>						
Female	36	25	61			
Male	60	40	100			
Total	96	65	161	0	1	1.00
<b>Child age</b>						
Preschool	27	24	51			
Primary	42	30	72			
Secondary	24	14	38			
Total	93	68	161	0.75	2	0.69
<b>Parent relationship</b>						
Mother	75	22	97			
Father	20	10	30			
Total	95	32	127	0.90	1	0.34
Note: Totals vary due to missing data						

Reasons given by parents for choosing not to participate or withdrawing participation included one parent who wanted to wait until a later stage of their child's treatment, two parents whose children did not have cancer<sup>3</sup>, one parent whose child's treatment had finished, and one parent who did not have sufficient English language to complete the survey. The researcher's reflective journal noted anecdotally that parents occasionally commented that their child did not experience any pain. Due to the anonymity of the survey it cannot be established whether these parents participated.

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<sup>3</sup> Both children had aplastic anaemia, a condition which is treated in a similar manner to cancer

### ***6.2.3 Demographic description of the survey sample***

#### **6.2.3.1 Child demographics**

Table 6.2 displays child demographics for the survey sample. The most frequent diagnosis was leukaemia and most frequent ethnicity was white. There were more males than females and children had most frequently been diagnosed less than six months prior to recruitment. Note that all children were on active treatment, so although relapse data were not collected, it can be assumed that those who had been diagnosed 5+ years ago were children who had relapsed. A range of ages were represented.

Table 6.2: Child demographics

<b>Demographic data (child)</b>		<b>n (%)</b>
Age	1	4 (4)
	2	4 (4)
	3	9 (8.9)
	4	10 (9.9)
	5	15 (14.9)
	6	4 (4)
	7	7 (6.9)
	8	5 (5)
	9	7 (6.9)
	10	4 (4)
	11	6 (5.9)
	12	4 (4)
	13	3 (3)
	14	2 (2)
	15	4 (4)
	16	5 (5)
Gender	Female	36 (35.6)
	Male	60 (59.4)
Ethnicity	Asian	11 (10.9)
	Black	8 (7.9)
	Mixed	12 (11.9)
	Other	2 (2)
	White	65 (64.4)
Diagnosis	Leukaemia	56 (55.4)
	Lymphoma	10 (9.9)
	Brain	11 (10.9)
	Solid tumours	16 (15.8)
	Other	2 (2)
Time since diagnosis	0-6 months	44 (43.6)
	6-12 months	9 (8.9)
	1-3 years	35 (34.7)
	3-5 years	8 (7.9)
	5+ years	2 (2)
Time since diagnosis (pre/post 6 months)	Pre- 6 months	44 (43.6)
	Post- 6 months	54 (53.4)
Note: totals vary due to missing data		

## Parent demographics

Table 6.3 displays demographic data for parents of the survey sample. The most frequent age range was 35-44 years, the most frequent ethnicity was white, and most frequent relationship was mother. A range of educational backgrounds and incomes were represented.

Table 6.3: Parent demographics

Demographic data (parent)		n (%)
<b>Relationship</b>	Mother	75 (74.3)
	Father	20 (19.8)
	Other	2 (2)
<b>Parent age</b>	25-34	22 (21.8)
	35-44	53 (52.5)
	45-64	23 (22.8)
<b>Parent ethnicity</b>	White	71 (70.3)
	Asian	11 (10.9)
	Black	12 (11.9)
	Mixed and Other	4 (4)
<b>Income per year</b>	Less than £14,000	13 (12.9)
	£15,000 – £24,000	25 (24.8)
	£25,000 – £39,000	14 (13.9)
	£40,000 – £59,000	11 (10.9)
	More than £60,000	26 (25.7)
<b>Education</b>	Didn't finish school	22 (21.8)
	Finished school	12 (11.9)
	Certificate or partial studies	43 (42.6)
	Completed a bachelor's degree	16 (15.8)
	Completed a postgraduate degree	2 (2)
Note: totals vary due to missing data		

#### **6.2.4 Descriptive statistics for survey results**

There was a statistically significant correlation between MAQ and PPEP scores ( $\rho=.405$ ,  $p<.001$ ). Figure 6.1 displays percentage response for each item of the PPEP. When strongly agree, agree and slightly agree were combined, highest agreement (86%) was for the statement “*children in pain have trouble sleeping*”. Lowest agreement (24%) was for the statement “*children feel less pain than adults*”. The inverse is true when strongly disagree, disagree and slightly disagree were combined. Highest uncertainty (15%) was for the statement “*children exaggerate pain*”, and lowest uncertainty (1%) was for the statements “*children always express pain by crying or whining*” and “*children who are playing are not in pain*”.

Figure 6.2 displays percentage responses for each MAQ item. When strongly agree, agree, and slightly agree were combined, highest agreement (73%) was for the statement “*side-effects are something to worry about when giving children pain medication*”. Lowest agreement (28%) was for the statement “*using pain medication for children’s pain leads to later drug abuse*”. The inverse is true when strongly disagree, disagree, and slightly disagree were combined. Highest uncertainty (33%) was for the statements “*it is unlikely a child will become addicted to pain medication if taken for pain*” and “*giving children pain medication for pain teaches proper use of drugs*”. Lowest uncertainty (9%) was for the statement “*pain medication works best if saved for when the pain is quite bad*”.

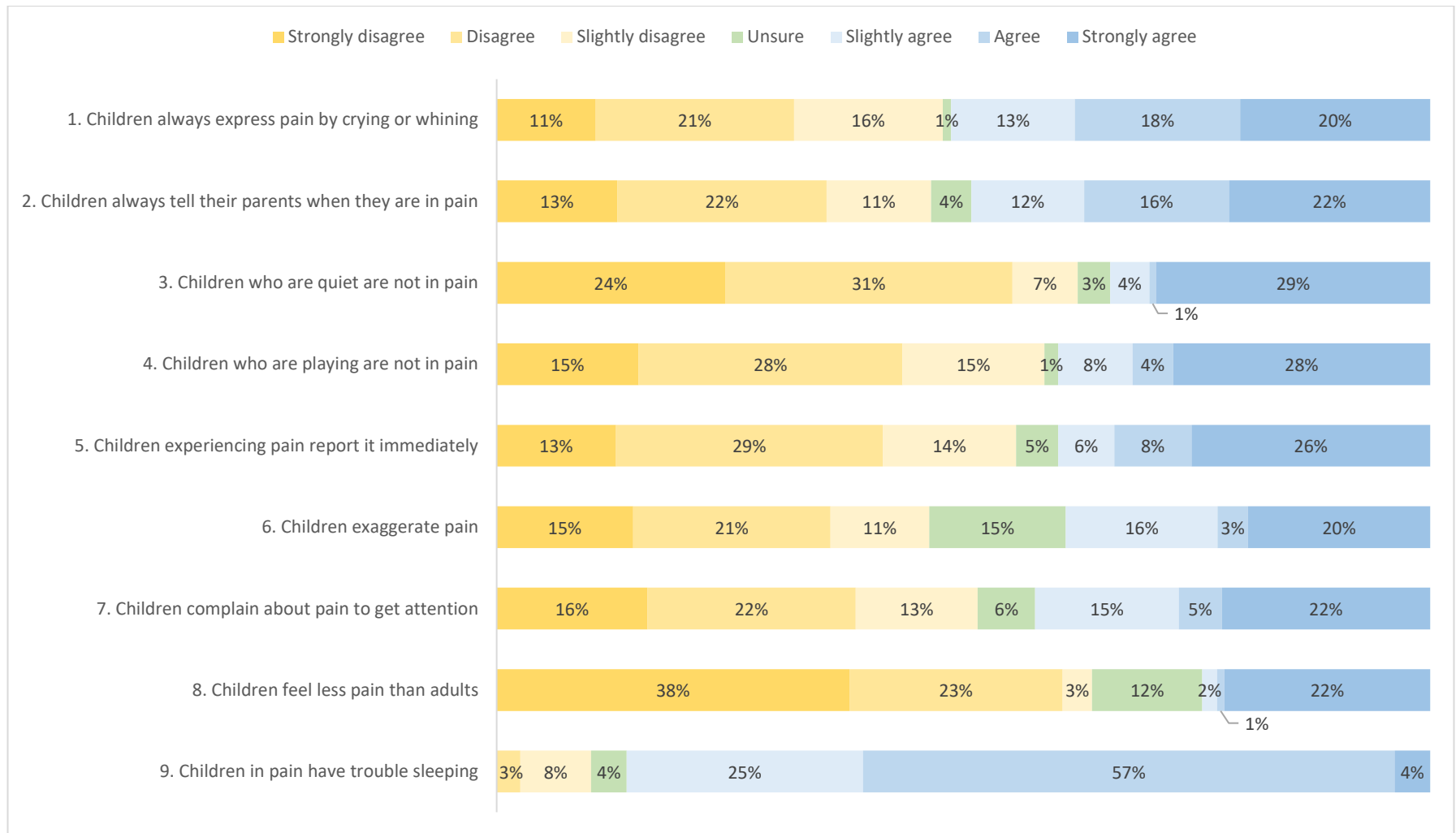


Figure 6.1: Percentage response for PPEP items

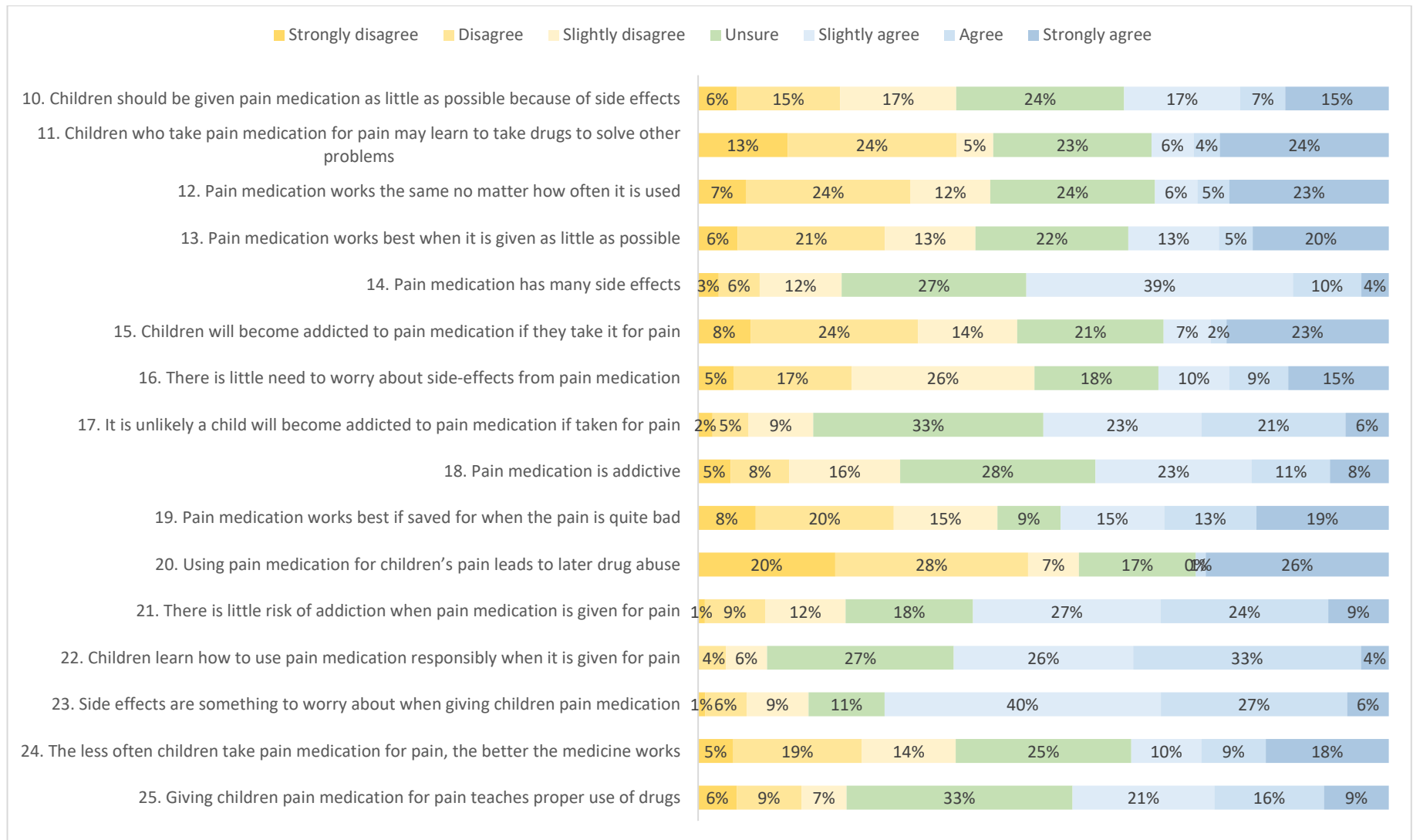


Figure 6.2: Percentage response for MAQ items

### 6.2.5 Comparison of demographic data with scales and sub-scales

Table 6.4 displays results of inferential statistics used to analyse child and parent factors as they related to scales and sub-scales. A small negative correlation was found between parent age and MAQ but not PPEP (Cohen and Holiday, 1982; Penn *et al.*, 2008). A statistically significant difference was found when ethnicity was compared to both scales and two sub-scales.

Table 6.4: Inferential statistics tests and significance

Independent variable	Dependent variable	Statistical Test	Significance
Time since diagnosis (categorical, 5 categories)	PPEP PPEP active loud PPEP quiet inactive PPEP attention seeking MAQ MAQ avoidance MAQ appropriate use MAQ fear of side-effects	Kruskal-Wallis	p=.949 p=.638 p=.362 p=.199 p=.548 p=.552 p=.589 p=.121
Pre- and post-six months since diagnosis (categorical, 2 categories)	PPEP MAQ	Student's t-test	p=.660 p=.599
Diagnosis (categorical, 5 categories)	PPEP PPEP active loud PPEP quiet inactive PPEP attention seeking MAQ MAQ avoidance MAQ appropriate use MAQ fear of side-effects	Kruskal-Wallis	p=.729 p=.097 p=.396 p=.802 p=.597 p=.521 p=.521 p=.945
Child gender (categorical, 2 categories)	PPEP MAQ	Student's t-test	p=.773 p=.085
Child age (continuous)	PPEP MAQ	Pearson's correlation	p=.065 p=.179
Parent age (categorical, 4 categories)	PPEP MAQ	Spearman's rho	p=.079 r= -.21, p=.047
Parent ethnicity (categorical, 5 categories)	PPEP	Kruskal-Wallis	Asian and White (h=24.2, p=.037)
	PPEP active loud		Asian and White (h=31.5, p=.004)
	MAQ		Asian and White (h=25.2, p=.015) Asian and other (h=40.5, p=.039)
	MAQ avoidance		Asian and White (h=27.8, p=.01) Asian and other (h=42.2, p=.048)
	PPEP quiet inactive PPEP attention seeking MAQ appropriate use MAQ fear of side-effects		p=.143 p=.694 p=.188 p=.070



### **6.3 Pain diary findings**

Results of the pain diary analysis are described below. Initially investigations into data distribution are presented, followed by results of participant attrition analysis. Descriptive statistics were used to provide information on the sample, and a combination of descriptive and inferential statistics were used to analyse pain diary results. Finally, the results of qualitative pain diary data are presented.

#### ***6.3.1 Data quality and distribution***

Data quality for pain diaries was low with many fields missing data and many days incomplete. Some fields, such as outcome, had insufficient data to analyse statistically. Pain score was completed for 82% of episodes. Distribution investigations reveal pain score for episode data and mean pain score for aggregate data to be negatively skewed. Non-parametric tests were used to analyse these variables. Maximum pain score for aggregate data were normally distributed.

#### ***6.3.2 Participant attrition***

Pain diaries were received from 37/101 (36.6%) potential participants. Table 6.5 displays results of a chi-square calculation to ascertain whether there were any statistically significant demographic differences between survey and pain diary sample. The purpose was to ascertain whether the data collection method had skewed the sample demographically. Five participants did not provide relationship and child gender information, and a further three participants did not provide information about their child's age so have been excluded from the chi-square calculation. One set of parents completed the pain diary together and were also excluded from the chi-square calculation because this would have resulted in cells which were too small to analyse. No statistically significant difference was found in gender and age of child, or parent relationship depending on whether parents chose to complete the pain diary or to complete only the survey.

Table 6.5: Screening demographics and chi-square calculation

	Completed pain diary & survey	Completed survey only	Total potential participants	Chi-square	df	p-value
<b>Child gender</b>						
Female	14	22	36			
Male	23	37	60			
Total	37	59	96	<.001	1	.957
<b>Child age</b>						
Preschool	12	15	27			
Primary	15	27	42			
Secondary	9	15	24			
Total	36	57	93	.54	2	.76
<b>Parent relationship</b>						
Mother	29	46	75			
Father	6	14	20			
Total	35	60	95	.51	1	.475
Note: Totals vary due to missing data						

### 6.3.3 Demographic description of pain diary sample

#### 6.3.3.1 Child demographics

Table 6.6 displays child demographics for the pain diary sample. The most frequent diagnosis was leukaemia and most frequent ethnicity was white. There were more males than females and most children had been diagnosed for less than six months at point of recruitment. A range of ages were represented.

#### 6.3.3.2 Parent demographics

Table 6.7 displays demographic data for parents of the pain diary sample. The most frequent age range was 35-44 years, the most frequent ethnicity was white and most frequent relationship was mother. A range of educational backgrounds and incomes were represented.

Table 6.6: Child demographics

<b>Demographic</b>	<b>Participant response</b>	<b>n (%)</b>
Age	1-3	8 (21.6)
	4-7	14 (37.8)
	8-12	13 (40.5)
Gender	Female	14 (37.8)
Ethnicity	White	27 (73.0)
	Mixed	4 (10.8)
	Asian	3 (8.1)
	Black	2 (5.4)
Diagnosis	Leukaemia	22 (59.5)
	Lymphoma	5 (13.5)
	Solid tumours	7(18.9)
	Brain	2 (5.4)
Time since diagnosis	0-6 months	23 (62.2)
	6-12 months	2 (5.4)
	1-3 years	8 (21.6)
	3-5+ years	4 (10.8)
Note: Totals vary due to missing data		

Table 6.7: Parent demographics

<b>Demographic</b>	<b>Variable</b>	<b>n (%)</b>
Relationship	Mother	29 (78.4)
	Father	6 (16.2)
	Other	2 (5.4)
Parent age	25-34	5 (13.5)
	35-44	22 (59.5)
	45-64	10 (27)
Parent ethnicity	White	29 (78.4)
	Asian	3 (8.1)
	Black	4 (10.8)
Income per year	Less than £14,000	3 (8.1)
	£15,000 – £24,000	2 (5.4)
	£25,000 – £39,000	13 (35.1)
	£40,000 – £59,000	7 (18.9)
	More than £60,000	10 (27.0)
Education	Finished school	2 (5.4)
	Certificate or partial studies	15 (40.5)
	Completed a bachelor's degree	10 (27.0)
	Completed a postgraduate degree	9 (24.3)
Note: Totals vary due to missing data		

### 6.3.4 Pain score

A pain score was recorded for 1769/2137 (82.8%) episodes. Of these episodes, 465 (26%) recorded a pain score of one or more. In 292 episodes (17%), clinically significant pain of three or more on the NRS (Fortier *et al.*, 2014) was recorded. Pain score frequencies are displayed in Figure 6.3.

Only six children did not have clinically significant pain on at least one occasion during the one-month pain diary period. Three children had a pain score of zero for the duration of the pain diary. Twenty-nine children had three or more episodes of clinically significant pain during the pain diary period. One child had clinically significant pain for 90% of the pain diary period, and one child had clinically significant pain for 100% of the pain diary period.

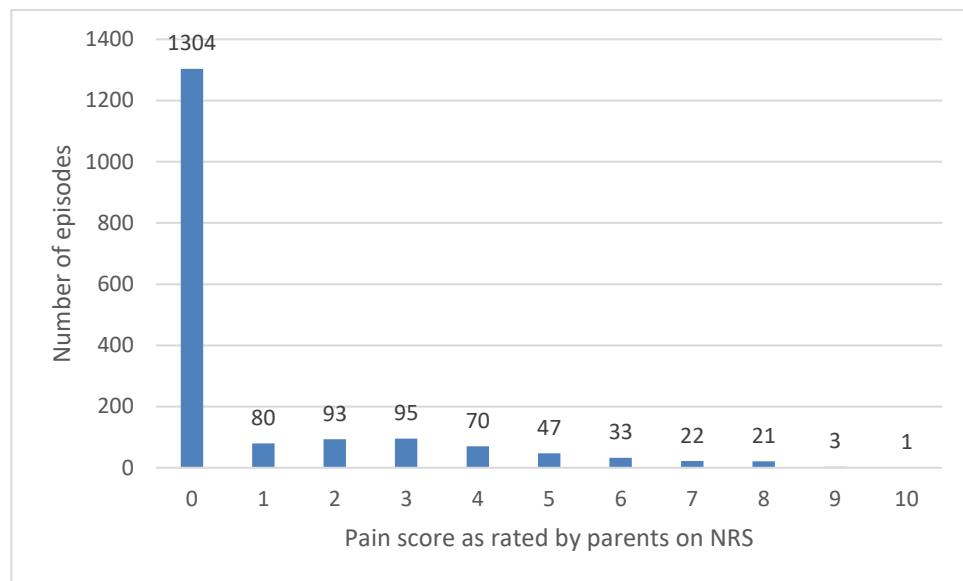


Figure 6.3: Frequency of pain scores as reported by parents on NRS

Table 6.8 displays statistical comparisons of pain scores to demographic data. There was no statistically significant difference detected when comparing pain score to diagnosis (one-way ANOVA), time since diagnosis (one-way ANOVA) and pre-and post-six months since diagnosis (Student's T-test). Rationale for choice of inferential statistics is described in Sections 5.5.4.5 and 5.6.4.

Table 6.8: Comparison of pain scores to child demographics

Independent variable	Dependent variable	Statistical Test	p-value
Diagnosis	Maximum pain	ANOVA	0.147
	Mean pain	Kruskal-Wallis	0.436
Time since diagnosis	Maximum pain	ANOVA	0.799
	Mean pain	Kruskal-Wallis	1.000
Pre- and post-six months since diagnosis	Maximum pain	Student's t-test	0.818
	Mean pain	Mann-Whitney U	0.925

### 6.3.5 Location of pain

Pain location was recorded for 492 episodes. In 36 of these episodes no pain score was recorded, in one episode pain was recorded as zero, and in two episodes pain was recorded as “?”. Table 6.9 displays frequency of pain locations for episode data and aggregated data. In 59 episodes, pain was reported in more than one location which meant intensity of pain for each location could not be calculated. For children who had a multimodal distribution of pain locations, the most frequent locations recorded by their parents have all been counted. Three participants had no pain for the pain diary duration and so did not record a pain location. Seventy-two percent of arm pain can be attributed to one child who had 23 episodes of arm pain.

Table 6.9: Frequency of pain locations

<b>Location</b>	<b>Number (%) of episodes</b>	<b>Number (%) of children for whom this is the most frequent pain location</b>
Abdomen	123 (25)	11 (26)
Legs	104 (21)	11 (26)
Mouth / Throat	99 (20)	4 (9)
Head	71 (14)	5 (12)
Bottom	66 (13)	1 (2)
Arms	32 (7)	1 (2)
Back	30 (6)	4 (9)
Other	21 (4)	3 (7)
Don't know	15 (3)	1 (2)
Chest	10 (2)	2 (5)

### 6.3.6 Cause of pain

A cause was recorded for 448 pain episodes. Table 6.10 displays frequency of causes of pain for episode data and aggregated data per child. In 60 episodes, pain was reported to have more than one cause which meant intensity of pain per cause could not be calculated. For children who had a multimodal distribution of cause of pain, the most frequent causes recorded by their parents have all been counted. Four participants did not record a pain cause for the duration of the pain diary. Of these, three did not record pain for the duration of the pain diary. All pain from disease can be attributed to one child, 50% of pain from procedures can be attributed to one child who had five episodes of procedure pain, and 56% of pain from infection can be attributed to one child who had 14 episodes of infection pain. Other causes of pain included specific daily activities like carrying a school bag, and avascular necrosis.

Table 6.10: Frequency of causes of pain

Cause	Number (%) of episodes	Number (%) of children for whom this is the most frequent cause of pain
Chemo-toxicity	120 (23)	9 (23)
Constipation / diarrhoea	90 (17)	7 (18)
Other	62 (12)	3 (3)
Mucositis	60 (12)	2 (5)
Drug side-effects	46 (9)	3 (8)
Don't know	27 (5)	4 (10)
Unwell / infection	25 (5)	2 (5)
Surgery	25 (5)	2 (5)
Being active	25 (5)	3 (8)
Nausea	23 (4)	2 (5)
Procedure	10 (2)	2 (5)
Disease	4 (1)	1 (3)

### 6.3.7 Response to pain

A parent response to pain was recorded for 506 episodes. In 121 episodes, parents recorded both a pharmacological and a non-pharmacological response. No action was taken for 51 episodes. At aggregate level, there were no children for whom “no action” was the most frequent response to their pain. Three participants did not record any pain for the pain diary duration so did not record a response either. A univariate ANOVA revealed that as pain scores increased, there was a statistically significant increase in parents’ simultaneous administration of pharmacological and non-pharmacological responses in combination when compared to only administering pharmacological interventions (mean difference=1.48,  $p<.0001$ ), only using non-pharmacological interventions (mean difference=1.30,  $p<.0001$ ), and no action (mean difference=1.02,  $p=.009$ ). All other interactions were not statistically significant.

#### 6.3.7.1 Pharmacological interventions administered

Parents administered pharmacological interventions in response to 231 episodes. This was the most common response for 10 children (26%). Figure 6.4 displays the number of pharmacological interventions received by children over the pain diary period. Most children (56.7%) received fewer than four pharmacological interventions throughout the one-month pain diary period. Figure 6.5 displays the number of analgesic drugs administered in response to varying levels of pain. For

20 episodes, one or more analgesic drugs were administered when no pain score was recorded. These were all by one participant who used the comments section to note that being asked for a pain score irritated the child and therefore none was recorded. On every occasion, when the pain score was recorded as zero, no analgesic drug was administered. Children received an analgesic drug in 21% of episodes where a pain score was recorded as one or more. In 71% of episodes where children had clinically significant pain, no analgesic drug administration was recorded.

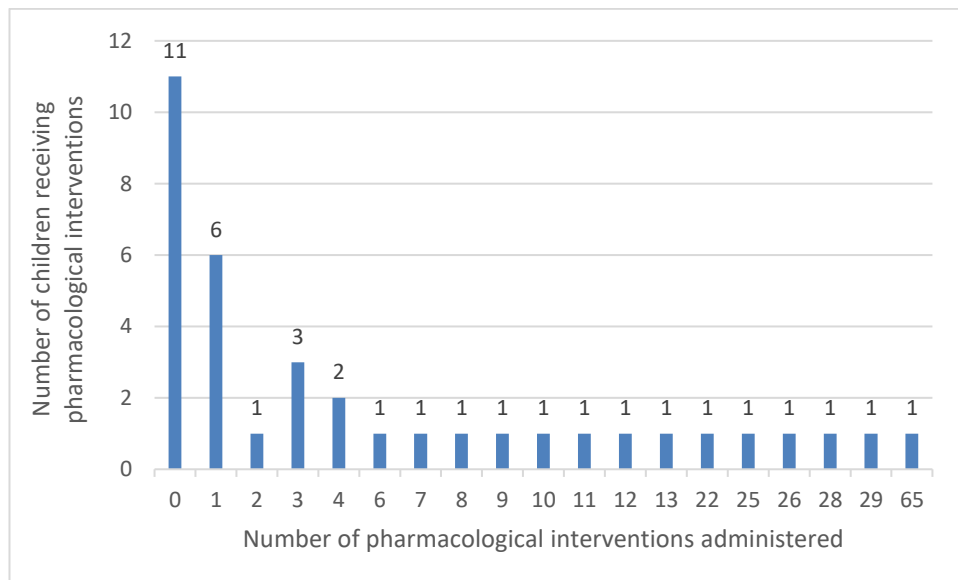


Figure 6.4: Number of pharmacological interventions per child

Table 6.11 displays the most frequent response for each type of pharmacological intervention and each type of analgesic drug for episode data (number of episodes) and aggregate data (number of children for which this was the most frequent response). All co-codamol administration can be attributed to one child, and all codeine<sup>4</sup> and pregabalin administration can be attributed to another child. Dose appropriate frequencies could not be calculated for the majority of pharmacological interventions due to doses being provided in millilitres rather than milligrams or the British Nursing Formulary for children providing dosing dependent on a child’s weight which was not collected (Paediatric Formulary Committee, 2016).

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<sup>4</sup> Codeine is no longer recommended for use in children (Andrzejowski and Carroll, 2016; Cheng and Tattermusch, 2014; Hanmod and Gera, 2016; MHRA, 2013)

Table 6.11: Frequency pharmacological intervention and type of analgesic drug administered

	Number (%) of episodes	Number (%) of children for whom this is the most frequent response
<b>Type of pharmacological intervention administered</b>		
Analgesic drug	119 (50)	14 (50)
Topical	59 (25)	6 (21)
Antiemetic	29 (12)	4 (14)
Laxative	26 (11)	3 (11)
Antidiarrheal	3 (1)	1 (4)
<b>Type of analgesic drug administered</b>		
Paracetamol	78 (51)	12 (71)
Morphine	53 (35)	5 (29)
Pregabalin	18 (12)	0 (0)
Co-codamol	3 (2)	0 (0)
Codeine	1 (1)	0 (0)

### 6.3.7.2 Non-pharmacological intervention responses

A non-pharmacological intervention response was recorded for 345 (55%) episodes. This was the most common response for 28 children (74%). Table 6.12 displays frequency of non-pharmacological intervention responses for episode and aggregated data. In 44 of these episodes, parents recorded more than one non-pharmacological intervention response. For participants who had a multimodal distribution of response, their most frequent responses have all been counted. One child received 50% of oral care recorded. Responses classified as “other” included life adjustments such as arranging for the child to be carried, applying topical cream, and adjusting the child’s environment.



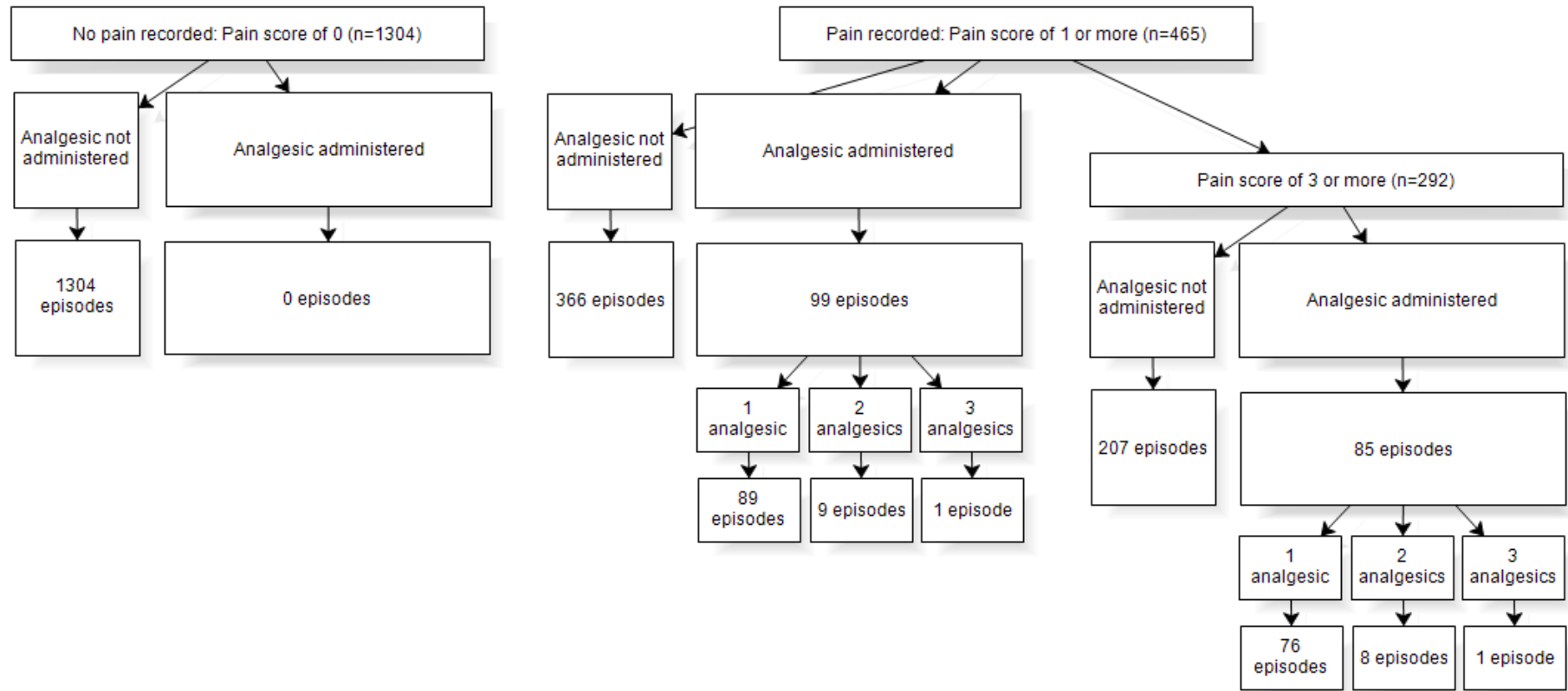


Figure 6.5: Pharmacological interventions in response to pain score

Table 6.12: Frequency of non-pharmacological intervention responses

<b>Non-pharmacological intervention response</b>	<b>Number (%) of episodes</b>	<b>Number (%) of children for whom this is the most frequent response to pain</b>
Cuddles	86 (17)	6 (16)
Food and Drink	67 (13)	3 (8)
Distraction	65 (13)	3 (8)
Massage	62 (12)	7 (19)
Sleep / rest	45 (4)	4 (11)
Heat	29 (6)	3 (8)
Bath / shower	28 (5)	2 (5)
Oral care	26 (5)	1 (3)
Verbal reassurance / comfort	18 (4)	3 (8)
Toilet	18 (4)	0 (0)
Exercise	12 (2)	1 (3)
Hospital or contact HCP	8 (2)	1 (3)
Other	48 (9)	3 (8)

### **6.3.8 Pain episode outcomes**

For 378 episodes, parents provided data on the outcome of their intervention. Due to this field being obtained as a result of quantising qualitative data into quantitative data (Section 5.6.4), it would not have been appropriate to conduct inferential statistics with this data. Figure 6.6 displays outcome data according to improvement or no improvement in pain. For 27 children, improvement in pain was the most frequent outcome. For six children, no improvement in pain was the most frequent outcome. Four children had no data provided by their parents on the outcome of the pain episode for the entire pain diary period.

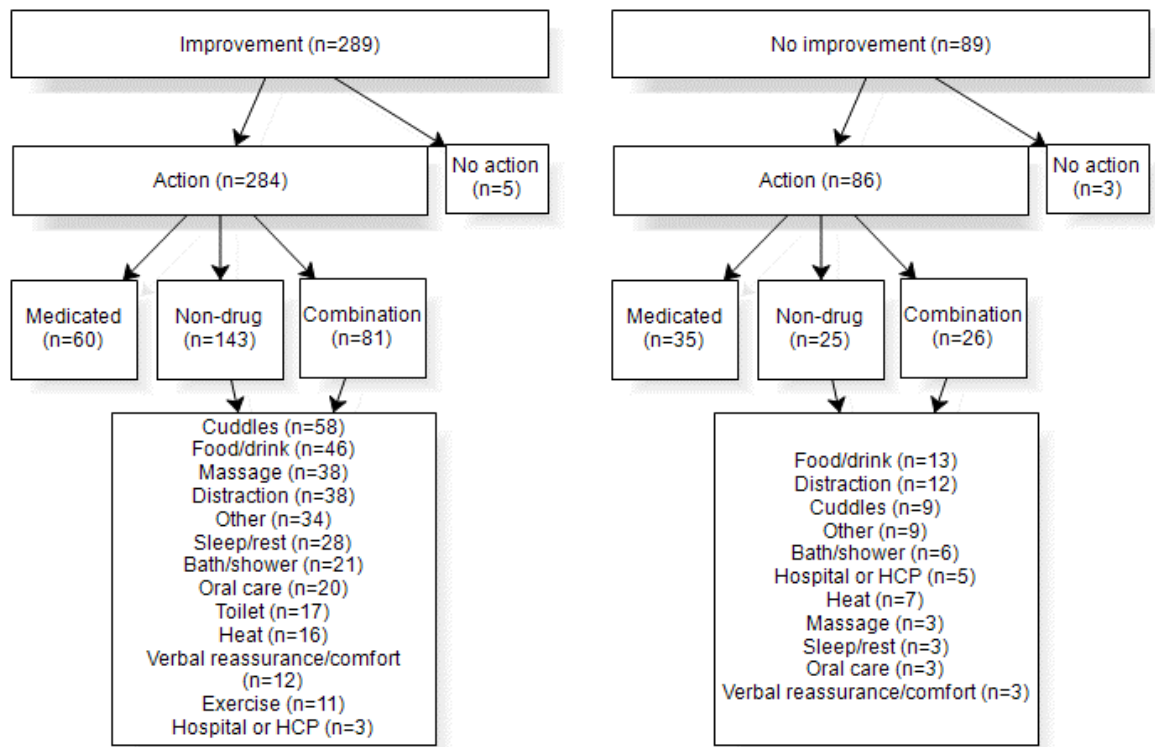


Figure 6.6: Outcome of episode and parent response

### 6.3.9 Reason for no action

Participants provided a total of 40 comments under “If no action is taken or required, please say why”. Many of the 313 comments under “Any other comments” also related to why no action had been taken so these two fields were analysed together. Due to the brevity of responses, analytic approaches such as thematic analysis would not have been appropriate to analyse this data. The results of a qualitative content analysis examining manifest content with low abstraction degree and low interpretation level (Graneheim and Lundman, 2004) is displayed in Table 6.13. Comments covered a broad array of topics which were separated into comments providing context and comments relating to reasons parents had taken no action. Contextual comments included comments on the practicalities of living with a child with cancer such as frequent hospital admissions and chemotherapy administrations. Parents commented on symptoms such as nausea, lethargy, and mood swings. Parents also made several positive comments regarding lack of pain or appreciation for HCP care. Reasons for no action were coded into four categories and nine subcategories. Numbers and percentage of participants who commented on each category are provided in Table 6.13.

Table 6.13: Reasons for no action in response to pain

Category	Subcategory	Exemplar	Number (%) of participants who commented
Features of the pain	Pain not severe enough	<i>“Pain was bearable.”</i>	8(21.6)
	Pain went away	<i>“Nothing as pain went as quickly as it came. Pain already gone.”</i>	4(10.8)
Inadequacy of analgesic drugs	Child refusing pharmacological intervention	<i>“No medication given as she won't take it.”</i>	6(16.2)
	Side-effects of analgesic drugs	<i>“Pain killers will mask the fever or other symptoms of potential infection.”</i>	2(5.4)
	Analgesic drug won't help	<i>“He just needs to poo, and this is the only thing which cause and ease the pain.”</i>	2(5.4)
HCP influence	HCP advises against analgesic drug	<i>“Advised by doctors not to administer anything.”</i>	4(10.8)
	Waiting to seek advice from HCP	<i>“Waiting for nurse visit, will seek advice.”</i>	3(8.1)
	Not allowed to give analgesic drug	<i>“Not allowed to give Calpol.”</i>	2(5.4)
Daily challenges	Circumstances prevent analgesic drug administration	<i>“We were in the car.”</i>	3(8.1)

## **6.4 Interview findings**

This section provides a comprehensive and representative overview of interview findings. Initially a description of participants is provided, followed by an overview of the analysis processes.

Interview findings are organised into a model of parents' management of children's cancer pain at home which is presented first in diagrammatic form, then as a written summary, and then in more detail.

### ***6.4.1 Participant description***

Description of participants and their children is provided in Table 6.14. There were an equal number of boys and girls, and an equal distribution of children across three age groups. Children's ages ranged from 1-16 years (median five years old). Older children who had been diagnosed less than six months prior to recruitment were under-represented and older children who had been diagnosed more than six months prior to recruitment were over-represented. All other groups were recruited as per sampling framework in Table 5.3.

Table 6.14: Details of interview participants and location

<b>Time since diagnosis</b>	<b>Child pseudonym</b>	<b>Parent pseudonym</b>	<b>Parent gender</b>	<b>Child gender</b>	<b>Diagnosis</b>	<b>Child age group</b>	<b>Location of interview</b>	<b>Duration (minutes)</b>	<b>PPEP score</b>	<b>MAQ score</b>
<b>Less than six months</b>	Filip	Natalia	Mother	M	Leukaemia	Youngest	Café	70	3.3	4.1
	Ruby	David	Father	F	Wilm's tumour	Youngest	Hospital	75	4.0	3.8
	Annabelle	Stacey	Mother	F	Leukaemia	Youngest	Hospital	90	2.9	4.1
	Jessie	Jackie	Mother	F	Lymphoma	Middle	Hospital	111	2.1	2.6
	Justin	Angie	Mother	M	Leukaemia	Middle	Hospital	90	3.1	3.9
	Peter	Laura	Mother	M	Leukaemia	Middle	Hospital	75	2.4	3.5
	Pippa	Beth	Mother	F	Leukaemia	Oldest	Home	79	2.2	2.6
	Emma	Ruth	Mother	F	Lymphoma	Oldest	Café	56	2.8	2.9
<b>Greater than six months</b>	Michal	Jana	Mother	M	Neuroblastoma	Youngest	Hospital / home	103	2.3	3.5
	Dimitris	Elena	Mother	M	Neuroblastoma	Youngest	Hospital	54	2.9	3.8
	Alan	Helen	Mother	M	Leukaemia	Youngest	Telephone	65	5.4	4.5
	Eleanor	Suzannah	Mother	F	Leukaemia	Middle	Home	75	2.9	2.5
	Lucy	Lisa	Mother	F	Lymphoma	Middle	Telephone	56	3.7	2.0
	Poppy	Georgia	Mother	F	Leukaemia	Middle	Researcher office	95	3.9	3.8
	Ollie	Brenda	Mother	M	Leukaemia	Oldest	Telephone	84	2.9	2.9
	Raj	Priya	Mother	M	Brain tumour	Oldest	Hospital	44	2.9	4.6
	James	Pauline	Mother	M	Lymphoma	Oldest	Hospital	65	2.1	2.9
	Carrie	Margret	Mother	F	Leukaemia	Oldest	Home	71	1.7	3.3

### 6.4.2 Analysis process

Interview data analysis followed the six phases of thematic analysis (Braun and Clarke, 2006) outlined in Table 5.4. When reviewing themes, attention was given to analysing relationships between themes to ensure their accurate definition (Clarke *et al.*, 2015). Ongoing refinement and reviewing of themes was aided by the development of “maps” which helped describe how themes lived within the dataset (Clarke *et al.*, 2015; Willig, 2013). The model of parents’ management of children’s cancer pain at home described below did not emerge passively from the findings: the researcher was active in the process of constructing, refining, and honing the model. This refinement process included constantly questioning data in relation to how the model worked. Several iterations were created before reaching the format presented below. Each iteration was described to the supervisory team who collaborated and encouraged thinking about cases in which the model was accurate as well as deviant cases.

These techniques led to an understanding of themes as distinct phases which parents went through as they managed their child’s pain. Initially two “maps” of different aspects of pain management were created; one concerning the pain management context and one concerning interventions. With further questioning of the data and analysis in partnership with supervisors, these were combined into one linear model. After processing and reprocessing (Howitt and Cramer, 2010), the researcher recognised the cyclical nature of pain management for parents. This led to a complicated cyclical model including five phases with many sub-themes. There remained difficulties with relationships between themes, and inconsistencies between data and model. When it was recognised that in each phase (theme) there were situational factors and a parent response (subthemes), the model was simplified to four phases. Initially, “every child is different” was a sub-theme of the “context” phase, but this theme appeared to interact with many other themes and sub-themes and could not be incorporated neatly into any phase of the model. With further questioning of themes, examining relationships between themes, and constant comparison to the data, it became apparent that “every child is different” was a key concept threaded throughout the model affecting every phase. The resultant simplified model presents a comprehensive account of parents’ management of children’s cancer pain at home. Figure 6.7 displays this model, which is a way of depicting the findings as a whole diagrammatically.

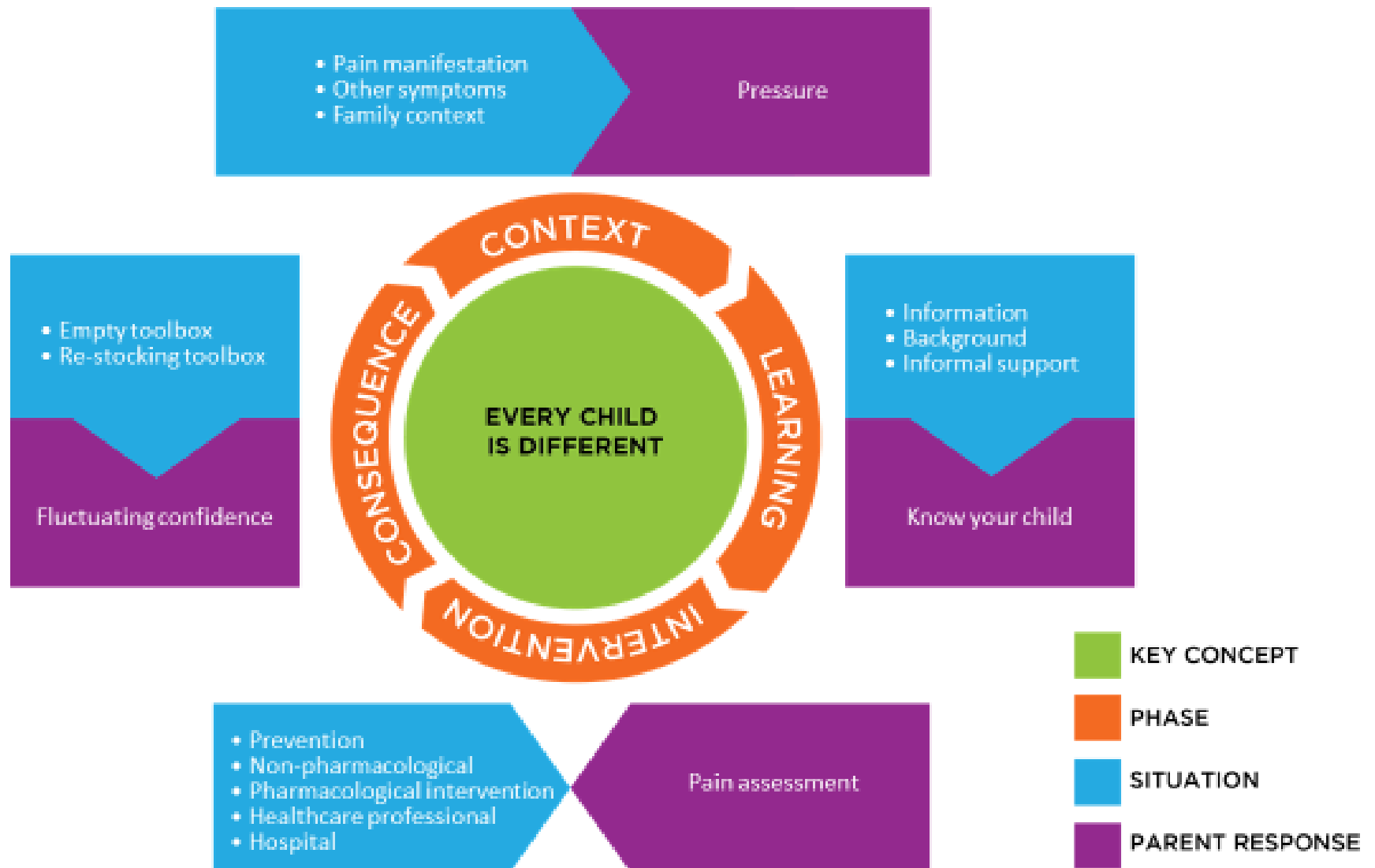


Figure 6.7: Diagrammatic representation of interview results



### ***6.4.3 Summary of parents' management of children's cancer pain at home model***

Findings showed parents' management of children's cancer pain at home as a sequential, cyclical process, repeating through four phases; context, learning, intervention, and consequence. Each phase has several situational factors and a parents' response. The key concept threaded throughout these four phases is that every child is different:

- **Context:** Parents described how the individuality of their child impacted on their response to the context. Each child's uniqueness contributed to parents' feelings of being alone and helpless; parents were acutely aware of their pain management responsibility which created a sense of pressure.
- **Learning:** Pressure regarding their pain management responsibility drove parents into a learning phase using information sources, personal background, and informal support. The purpose of this learning was to "know your child" which was considered key to successful pain management. Because every child is different, parents thought they were the only person able and willing to put effort into becoming an expert in their child's pain management.
- **Intervention:** The learning phase enabled parents to select an appropriate intervention for their child. Parents described five types of intervention available to them when their child was in pain: prevention, non-pharmacological interventions, pharmacological interventions, calling a HCP, and going to hospital. The parental response in this phase was pain assessment. The key concept of every child being different manifested in unique pain behaviours. Parents were able to assess their child's pain accurately and select an appropriate intervention.
- **Consequence:** Pain episodes could conclude positively or negatively. Many parents were dissatisfied with interventions available to them and at times felt their pain management "toolbox" was empty. Some parents were able to "re-stock" their "toolbox" with flexible and creative non-pharmacological interventions. Children responded differently to interventions and as a consequence, parents' confidence fluctuated. Generally, parents described increasing confidence over time, but some parents described pain episodes which caused their confidence to decrease or disappear entirely. The consequence of each pain episode created context for the next.

### ***6.4.4 Detail of parents' management of children's cancer pain at home model***

Parents' management of children's cancer pain is described in detail below. Initially the key theme of "every child is different" is described. Following this each phase is described first in terms of the situation and then parents' response. Quotes have been used to illustrate each phase. The key theme is threaded throughout to show how this theme interacts with sub-themes at each phase.

#### **6.4.4.1 Every child is different**

*“...every child is different. Every child is different. Every child responds differently” – Jana*

The concept that every child is different was key in parents' management of children's cancer pain at home. This concept is threaded throughout the model's four phases: context, learning, intervention, and consequence. The uniqueness of each child creates a context at home in which the parent is the only one responsible, alone in their pain management role, and feeling pressure as a result. Parents go through an essential phase of learning specifically about their child, to reach a point where they know their child. Every child expresses their pain differently, so parents have a unique ability to assess their child's pain and select an intervention. It is of note that “every child is different” does not mean outside of cancer settings every child is the same. Rather that this concept is key in creating challenges for parents' management of children's cancer pain. Parents felt that their child's response to treatments were unknown and could differ from other children with the same diagnosis and treatment. Once parents know their child they can accurately assess their individual pain responses and levels. The consequence is fluctuating confidence in their pain management role.

#### **6.4.4.2 Context**

Context includes situational factors of pain manifestation, the child's experience of other symptoms, and family response. Parents' response to context is the feeling of pressure.

##### **6.4.4.2.1 Pain manifestation**

Children experienced pain in many parts of their body, with a variety of causes. Parents frequently described “bottom” and abdominal pain. This pain was mostly due to constipation and occasionally due to diarrhoea. Less commonly, but still frequently, children experienced mouth and throat pain due to mucositis. Other locations included jaw, head, arms, legs, and back. Parents frequently attributed their child's pain to chemotherapy and steroids. Parents also described pain from procedures such as bone marrow aspirates, lumbar punctures, or intrathecal chemotherapy, which happened in hospital but remained painful on discharge. Other causes included muscle weakness, infection, and rashes. Parents recognised different types of pain, distinguishing between duration and intensity of pain:

*“...intermittent [pain], these are moments, this is not kind of chronic pain that continues...there's debilitating pain, and there's just I don't feel quite right today I'm under the weather sort of pain” – Jackie*

Parents reported that children experienced frequent low-level pain which resolved swiftly without intervention:

*“...he’ll just say, oh mummy my leg hurts, my hip hurts...it’s just a random pain...But normally it’s a passing thing where he’ll moan about it for a few minutes and then it’s gone.” – Angie*

Parents also described episodes of severe pain:

*“...that weekend when everything hurt...Her eyes were sort of gone in a way. She was suffering that day. So that was 10 [out of 10] yes.” – Ruth*

In addition to describing the presence of pain, parents also described the absence of pain:

*“...each drug has its own side-effect, but pain generally hasn’t been one of them” – Angie*

Parents were grateful for this and most referred to themselves and their child as “lucky”. Their perception of themselves as “lucky” was drawn from comparisons to other children and parents who they perceived had experienced more pain. They were not specific about which families made them feel lucky:

*“I think we’re quite lucky in the fact that she wasn’t in [pain], I know some children are in pain a lot and maybe if she was I would have dealt with things differently” – Jackie*

Parents often commented on age as both a positive and negative contextual factor. Parents of younger children described age as a barrier to helping children understand their pain, its cause, and how to resolve it. Below Suzannah described how she wished her child could understand the transient nature of her pain:

*“...for that child to understand that their pain is a time in their lives, it’s not forever, because children don’t have a concept [of] time” – Suzannah*

Parents of older children described their age as advantageous due to it providing their child with ability to reason:

*“...old enough to be able to reason with it. You’ve got to do it cos it’s going to make you better. So, he kind of does it.” – Angie*

Conversely, parents of older children worried about the psychological impact of pain on their child. One parent whose child had relapsed compared her child’s different psychological responses to pain with age:

*“...he’s a little bit older and he’s thinking about a lot more things...Whereas when they’re younger, because they don’t really know any better...he...coped a little bit better then, than he is now.” – Priya*

This potential for psychological damage caused by pain was recognised as a “blessing” in parents of younger children:

*“But it’s also been a blessing in terms of her not really being bogged down psychologically...An older kid would probably struggle with that a lot more. And so, the psychological benefit with being pre-school age...in the future she may not remember much or any of this which could be a blessing as well.” – David*

Pain also held psychological implications for parents. Many children experienced pain as a symptom of cancer prior to diagnosis, so when the child was in pain, parents feared their child may be relapsing:

*“...the leg pain I thought oh my god why can’t she cycle down the street? She could cycle down the street last week. Why can’t she do that this week? Oh my god could it be the leukaemia?” – Suzannah*

#### **6.4.4.2.2 Other symptoms**

Unprompted by the interview questions, parents discussed several other symptoms experienced by their children which interacted with pain. Nausea was a common example: As well as being distressing on its own, nausea interacts with pain through preventing children from wanting to, or being able to, take oral pain-relieving medications. Lisa describes how the solution to mouth pain caused by mucositis failed due to her child feeling nauseous:

*“the only thing we could give her was gelclair<sup>5</sup> which is a sachet of mouth rinse but Lucy at the time was feeling very sick and the taste and the strong aniseed taste made her feel even more sick, so she would rather not have that” – Lisa*

Nausea and taste changes affected nutrition, which in turn caused constipation which, as noted above, was often accompanied by pain as well as being a side-effect of opioid medication.

Conversely, pain from mucositis could limit nutritional intake:

*“...he didn’t want to eat obviously, couldn’t eat anything that was hard or sharp or what have you...he couldn’t it was hurting him to eat cos it was hard. So, it was a bit tricky trying to get him to eat much...So he’d lost quite a lot of weight over those few weeks” – Angie*

Another symptom which provides context in which children experienced pain at home was lethargy. Parents reported that their child lacked energy and spent lots of time sleeping and resting. When children did begin to mobilise, this could lead to pain:

*“And he’ll often say...my legs hurt...and often I think well he’s still building up his strength...being more active” – Helen*

Other symptoms included low blood sugar, and urinary retention.

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<sup>5</sup> Gelclair is an oral mouth wash which aims to relieve symptoms of mucositis.

#### **6.4.4.2.3 Family context**

Family context varied between participants. Some mothers reported occasions when their partner had been unhelpful in pain management:

*“...my husband is a brilliant businessman, he’s very confident in dealing with top people in the industry but he’s not as good when it comes to seeing someone in pain, or especially child[ren], so he was panicking, he was just making me even more, tense, more, making things worse instead of comforting me.” – Jana*

Some mothers reported an equal relationship with their partner:

*“I think we work collaboratively; we always would ring each other and say...what [do] you think?” – Laura*

Others reported their partner had a more positive input into their child’s pain management:

*“...my husband has been amazing, he’s been amazing. And he has a very practical approach to pain...He doesn’t panic at all” – Suzannah*

In most families, mothers were primarily responsible for pain management, often due to their partner’s work. In the quote below, this resulted in a lack of trust between parents in managing the child’s treatment:

*“...he’s kind of busy. Means that he hasn’t felt that confident around some of the treatment stuff...so I don’t always trust him to get it absolutely right, so I think in that sense I have felt quite lonely.” – Georgia*

Family context meant that for most participants responsibility for their child’s pain management fell to them alone. Their partner, whilst supportive, simply did not have the same exposure to their child and healthcare settings. The quote from Elena illustrates the key concept running throughout these findings and links it to the context. Each child has a different pain manifestation and a different experience of other symptoms:

*“Every child is different; every person is different. How our body reacts to things is going to be completely different from one person to the other.” – Elena*

#### **6.4.4.2.4 Parent response: Pressure**

Below Jana describes how the key concept of every child being different led to pressure on her. She articulates the family context and uniqueness of her child’s situation which led to “massive pressure”:

*“Mum is always left with everything on her shoulders...Dad has to work cos you have to survive somehow...such a massive pressure and it was so difficult cos I, I simply didn’t know. And even though you’ve got the support of people around you even though they educate you a lot, it’s down to you, cos you keep hearing the same thing, every patient is different, every child is different and that’s true...” – Jana*

This feeling of pressure was echoed by other parents. Georgia described a pain episode when Poppy’s father and grandparents were looking to her for answers:

*“And I suppose that comes back to the lonely thing and you...I have felt at times just huge pressure. And not feel like that pressure’s particularly shared and in one sense felt that pressure’s worse when people looked to me for the answer. And I’m like, I am as useless at this as all the rest of you, I know just as little...and you’re meant to pull the rabbit out of the hat and I’m like, I have no rabbit, I have no hats, right now I’d like to be anywhere but where I am.” – Georgia*

As suggested above, parents described feeling alone in their pain management role:

*“...every child is different so that’s why probably they [HCPs] leave the parents alone with the pain.” – Natalia*

Parents carried the burden of responsibility, not knowing what to do, and often felt helpless in crisis moments:

*“Well I guess it’s, whenever I’ve felt my most helpless feelings about Pippa’s cancer and health have been around pain. Just not knowing what to do.” – Beth*

#### **6.4.4.3 Learning**

Parents’ most consistent response to pressure was to become an expert. They put effort into learning about pain, cancer, and their new situation. It was an active choice, which this parent described as a coping mechanism:

*“That was my way of dealing with it, so I decided to learn more” – Jana*

Parents were aware of how their knowledge and lifestyle had changed over time. The quote below talks about clinical knowledge gained during treatment:

*“Cos it’s like a whole new, you have to, it’s a whole new knowledge base. I didn’t know anything about leukaemia, I didn’t know anything about the treatments. I just thought if you’re a kid and you’ve got cancer then you have chemotherapy, but I didn’t know what they were called, I didn’t know about blood tests, I didn’t know anything about that.” – Beth*

In addition to clinical knowledge, parents learned a new way of life; a “new normal”:

*“...we live in a different world now. Cos all things become normal now don’t they. But I guess you just find your own routine with things don’t you. And you get to learn what’s right, what’s wrong, what’s normal, what’s not normal for him.” – Angie*

Parents spoke about there being several iterations of re-learning about pain, cancer and their child as their child progressed through treatment:

*“But then of course it all changes with the next cycle of treatment. You’ve got to relearn all sorts of things.” – Suzannah*

Parents used information sources, personal background and informal support to learn and become an expert.

#### **6.4.4.3.1 Information**

Information sources included HCPs, online resources and parents' own instinct. Despite being "alone" in pain management at home, parents reported that HCPs could provide useful information which helped them in this role. Some parents reported being educated by HCPs about pain management whilst they were in hospital:

*"I think managing her pain at home was mainly the nurses [educating] ...before we left saying if this happens you need to try this" – Jackie*

Parents also observed pain management practices in hospital and used them to model pain management at home:

*"She associates hospital with play time...there's cupboards full of toys, the team are friendly, the whole thing is normalised for her...you can learn a lot. So, when you see that in hospital you can then take it home." - Suzannah*

The internet was a source of information which parents had conflicting views on. Some parents found the internet a useful resource and other parents felt strongly that it was not useful, led to negative emotional responses, or provided inaccurate information:

*"Google is...helpful cos you can find some information. But still you need to be very careful because you can find different, you can even find a potato diet for the leukaemia." – Natalia*

Although some parents were strongly opposed to the use of Facebook, it was often mentioned as a useful but limited source of information:

*"...sometimes you read [Facebook] and think 'why would you post that to a bunch of parents whose children have got cancer?', but if you take those with a pinch of salt it's a really good informative resource." – Laura*

For some parents, the extent of internet use changed over time:

*"I was very guarded, I did not use google...cos I just couldn't cope with it. And as time has gone on I have relaxed a bit...I feel more able to filter some of the information now, it's not going to send me down some horrible rabbit hole." – Georgia*

Instinct was another source of information for parents. Initially parents felt their trust in their instinct had been taken away. This may have been due to the trauma of their child having a cancer diagnosis. Over time, they realised instinct was important in their decision making:

*"...what I have learned though is to trust your instinct...And that's quite a hard thing to see sometimes in itself...generally that has tended to be correct and I've followed that." – Helen*

Despite access to several information sources, parents frequently expressed feeling that they had received insufficient information to manage pain at home. Parents wished they had known which pain management options were available to their child, advantages and disadvantages of each, how to assess their child's pain and what to expect at home. Parents suggested written or verbal provision of information as a potential solution:

*“... having someone that goes around the ward with all this information from all these different sources and having as a role...give you all the different options...it’s hugely needed...it would just help so much...Or if they won’t pay for someone to do it then some sort of information leaflet that is focused just on pain...something where parents have information, access to information.” – Jackie*

#### **6.4.4.3.2 Parents’ background**

Parents used different parts of their background including health, personality, profession, and parenting style to help them in their pain management role. Parents described using techniques they found helpful from past personal experiences of pain:

*“...it actually stems from myself because at the age of 12 I was diagnosed with rheumatoid arthritis...I’ve lived with pain quite a lot...So I used to sit with a hot water bottle...used to be quite soothing as well anyway. So, it’s mostly from my own personal experience” – Priya*

In addition, parents were aware that past painful experiences increased their empathy:

*“I mean obviously I believe because when I was younger I had that operation, you have that, you know that pain, that has obviously been with me ever since. So, whether that is subconsciously that knowledge just I know how much you can be in pain because it’s very painful.” – Pauline*

Parents reflected on ways in which their profession helped their pain management role.

*“The biggest issue we’ve had with Peter is taking his medication. So, and I’m a speech and language therapist my speciality was dysphagia, swallowing...I’ve worked trying to get medications into people who can’t swallow so I was like there must be a way, there must be a way.” – Laura*

Parents’ background helped them access the support they needed to manage their child’s pain.

*“I have a background in education...I’m used to communicating openly about things...so that’s helped enormously...to ensure that the pain management that we do at home and the care that we have at home is also matched by care at school...so I know where to access support certainly.” – Suzannah*

#### **6.4.4.3.3 Informal support**

Parents received support from family, friends, and other people with cancer experience. Family networks included grandparents, siblings and extended family. These individuals provided emotional and practical support and were trusted because they knew the parent and child.

*“Well just having another someone else here...Just talking through what I’m doing. Cos they obviously know Pippa really well as well...And then just giving me their opinion and it was someone who I respect...offloading on someone else just automatically helps I feel.” – Beth*

Other adults who had had cancer and friends who had clinical experience relevant to the cancer journey also provided practical support:



*“...because she had the cancer she had the chemotherapy, so she knew exactly how he’s feeling. And she said, leave him, if he doesn’t want to play, if he wants to lay down, he wants to lay down.” – Natalia*

Although parents received support from friends, most participants reported that unless a person had cancer experience, their ability to help was limited. Outside hospital, parents’ perception was that they were considered the unlucky parent whose child had cancer. One parent reported withdrawing from all her friends whose children did not have cancer:

*“I completely cut off my contact with friends who doesn’t have cancer in the family. Purely because it, for both of us, it was very uncomfortable...I really hate when people you know that mercy on their face and they say everything will be fine cos that annoys me cos you don’t know everything will be fine.” – Jana*

Many found support from other parents of children with cancer. Beth describes her relationship with the mother of another child with cancer who she met during a hospital admission. Beth recognises limitations in this mothers’ assistance because every child is different:

*“...she seems more knowledgeable about the whole thing anyway...if I had any questions...I would text her what do you think about this, what’s your opinion and she’d be really good. So, in terms of pain...so she would say this is what I’m doing with [my daughter]. Although [Pippa] was completely different and had different symptoms.” – Beth*

Parents were wary of support from other parents of children with cancer due to the potential emotional burden if the child died:

*“Obviously it’s a good thing to talk to other parents when everything’s going well. But then again when someone is less fortunate like [patient who died], that affect us a bit.” – Jana*

#### **6.4.4.3.4 Parent response: Know your child**

In the quote below, what Jackie alludes to is that because every child is different, no level of clinical expertise is enough if you do not know your child:

*“unless you’re an oncologist that has a child, but even then, you don’t know what works for them do you? I mean you might know more about their disease, but you don’t know what works for them...I think it’s just a process you learn.” – Jackie*

Parents were clear that the focus of this learning was to know their own child:

*“I’m not expert on neuroblastoma but definitely I’m expert on him.” – Jana*

In the next quote, Brenda describes how knowing her child related to pain management:

*“...you just learn what’s normal as in what to expect in the treatment and then what’s normal for your child...it’s day four and day five he always gets back pain...So I know or knew, when we’re on day four or five, don’t plan to walk to far, don’t plan to do anything energetic cos he’s likely to be in a lot of pain.” – Brenda*

Devotion and effort were evident in the way parents learned to know their child. Jackie describes observing her daughter, recording her observations, and the impact she felt this learning had on her pain management:

*“I think you have to put a lot of effort into working out your own child because I think that’s the main, the main thing to help you move forward with pain...I used to write a diary or jot things on my phone oh this happened this is how she reacted, just so that I knew for next time...So I did make a big effort to understand her and I think that’s probably the biggest key.” – Jackie*

By learning to know their child, parents were better able to intervene when their child was in pain.

#### **6.4.4.4 Intervention**

In this phase, the relationship between situation and parent response was two-way rather than linear. Parents described five interventions available when their child experienced pain. They are described below in the order of preference expressed by most parents.

##### **6.4.4.4.1 Prevention**

Parents initially preferred to prevent pain from occurring:

*“Wouldn’t it be awful if he woke up with a raging ulcer or something and you just think I could have prevented that (tearful)” – Laura*

Strategies included using mouthwash to prevent mucositis, preventing constipation by providing a high fibre diet or laxatives before symptoms arose, encouraging children to drink to prevent a headache, and encouraging children to eat frequent and small amounts to prevent abdominal pain. Strategies were personalised, and, as Suzannah described below, parents learned them as a result of effort put into knowing their child:

*“And as soon as you start to understand the side-effects of various drugs, you are able to then pre-empt the, some of the symptoms and alleviate them before they become really...I know that dexamethasone creates tummy ache for Eleanor...So I have written into her medical care plan at school and I have spoken to her school to say you must give her food on demand at this point...I think pre-empting things is very important.” – Suzannah*

##### **6.4.4.4.2 Non-pharmacological interventions**

Non-pharmacological interventions have been separated from pharmacological interventions to reflect the way in which parents talked about these interventions. Pharmacological interventions include paracetamol, ibuprofen, morphine, codeine, and drugs for neuropathic pain including pregabalin and gabapentin. Non-pharmacological interventions include physical strategies such as massage, psychological strategies such as distraction, non-pharmacological drugs such as

ondansetron, and other analgesic interventions such as topical analgesics. Negative attitudes expressed towards pharmacological interventions, which are described in the next section, meant parents generally preferred non-pharmacological interventions. Pharmacological interventions may have been the preferred choice prior to diagnosis, but since diagnosis, parents preferred non-pharmacological interventions and therefore non-pharmacological interventions are described first.

Parents expressed a belief that non-pharmacological interventions could be a sufficient solution:

*“...if you don’t have to take a medicine, then why would you, if there’s another solution?” – Laura*

Parents listed a wide variety of non-pharmacological interventions. These interventions included: heat, distraction, being there, reassurance, food, drink, cuddles and physical affection, herbal remedies, aromatherapies, massage, acupuncture, bath, treats, sleep, rest, going to the toilet, mouth washes, topical creams, numbing creams, laxatives, giving the child space and/or time, siblings, role play, hydrotherapy, TENS machine, fresh air, walking, mindfulness, phone apps, relaxation techniques, positioning, talking, placebo, plasters, probiotic yoghurts, singing, and physiotherapy.

Parents felt comfortable using more than one non-pharmacological intervention and were willing to try a variety of interventions simultaneously. In the quote below, Priya provides Raj with a massage, hot drink, hot water bottle, fresh air, a walk, cold drink, rest, and sleep for one pain episode:

*“...all of a sudden he said his front of his head was hurting, no first he said his head was hurting. So, I would sort of press it there for him and he says yeah that feels good and then I offered him some hot drink like tea...that made him feel better, but the pain wouldn’t go away and then...I gave him hot water bottle...and he felt a little bit better with it. And then I said to him look, you need to have some fresh air...let me put a chair here and...I said okay let’s go for a walk cos that fresh air and drinking water and then...why don’t you lie down and have a nap...and then he woke up and the headache was gone.” – Priya*

Parents viewed non-pharmacological interventions as “normal” in a way that pharmacological interventions were not:

*“I don’t always think that medicine’s the answer and I want Peter to get through this process in a normal fashion as possible...as opposed to going to the pharmacy and getting a medicine” – Laura*

#### **6.4.4.4.3 Pharmacological interventions**

If non-pharmacological interventions were ineffective, the next intervention available to parents in this study was to administer a pharmacological intervention. Deciding to administer a

pharmacological intervention was usually based on pain severity. Many parents did not frequently administer pharmacological interventions as they felt their child had only experienced what they described as low levels of pain:

*“...regularly and how often he was getting low levels of pain. And not necessarily enough to warrant giving [paracetamol]” – Brenda*

If severe, these parents said they would have chosen an analgesic drug:

*“I just wanted to make the point that obviously if I ever thought he needed it I would have given it to him. I’ve not withheld it.” – Laura*

Some parents held positive attitudes towards pharmacological interventions:

*“So, I think probably I’ve got a very relaxed attitude, rightly or wrongly to taking medicines...I will dose her up as much as she can have it and I won’t be worried.” – Beth*

When asked what advice they would give to other parents in a similar situation, some parents advocated administering pharmacological interventions:

*“Never be without [paracetamol], and definitely needing a bottle of [morphine] as a just in case.” – Brenda*

*“Don’t be afraid...a lot people don’t like using medicines. But at the end of the day, why make your child suffer? Just use medication, because it’s hopefully not gonna be for long.” – Ruth*

However, many parents had negative attitudes towards pharmacological interventions mostly fear of side-effects. Most discussion around pharmacological interventions focused on paracetamol or morphine, and parents held different attitudes towards each.

Parents expressed a range of attitudes towards paracetamol due to its antipyretic properties meaning administering it could mask a raised temperature in their child. At home, a raised temperature could be parents’ only sign of a potentially fatal infection in their immunocompromised child. Some parents expressed no hesitation in paracetamol administration while others were more cautious but would give it if their child had no sign of a raised temperature or were not neutropenic. Other parents never gave their child paracetamol due to fear it would mask a raised temperature:

*“Paracetamol definitely wasn’t allowed because of temperature spikes and stuff like that.” – Priya*

Receiving mixed messages from HCPs was an occasional barrier to pharmacological interventions. One parent had received conflicting information regarding paracetamol administration:

*“Some doctor in the [principle treatment centre], she said when he’s in pain give him paracetamol, she write me on a paper, how much I can give and without a temperature. But in [shared care] they said that I can’t give him anything. So that’s why was confusing...they give us antibiotics but nothing to kill the pain.” – Natalia*

Regarding morphine administration, one parent had encountered anti-morphine attitudes from HCPs:

*“I remember them saying she was morphine naive and they were quite pleased about that...they said it was a good thing.” – David*

When administering morphine some parents were aware of its pharmacological limitation of causing constipation as a side-effect. This was especially pertinent when their child had pain caused by constipation:

*“...the risk of constipation cos she is adamant she doesn’t want to take any laxatives so I’ve yet to get enough dose of morphine” – Margret*

Many parents held misconceptions based on personal experiences of exposure to morphine in the palliative phase of life, which influenced their attitude towards it. One parent had been exposed to morphine abuse within her family. In this excerpt, Beth bemoans a lack of guidance on morphine administration and whilst she admits negative attitudes towards morphine, she believes she is open to change:

*“...my husband had issues with it, with morphine, and his mum as well...So I’m quite frightened of morphine...even though it shouldn’t be subjective, it is...seeing people high on morphine when they shouldn’t have been when they’ve abused it...influenced my response when I saw this bottle of morphine in the bag.... So that was definitely a no go for me. She’s not having morphine. If someone had said to me, ‘well it would really help her, she’s in pain’, if her pain had been higher and a nurse or someone had said ‘look, you’ve been given morphine and it will help her as long as you stick to the guideline’ that we didn’t get. That would have been fine. I think I would have changed my mind about it. So, it’s not like I’ve got this really stuck attitude.” – Beth*

One child had a chronic pain condition as a result of cancer treatment for which she found paracetamol to be ineffective. She would likely be taking analgesic drugs for the rest of her life and her mother expressed worries about lifelong morphine administration:

*“You get used to one dose and then you up it and you up it and you up it. And if she’s got this for life, at what point do you say she’s got to cope with this pain rather than she’s going to be on morphine for the rest of her life?” – Margret*

Some parents also talked about not being allowed to administer ibuprofen, although compared to paracetamol, they were less clear on the reasons for this:

*“And he can’t have ibuprofen I don’t think. I can’t remember why but I don’t think he can.” – Angie*

Parents hesitated to administer pharmacological interventions due to believing their child was already “pumped full” of medication. Two distinctions were made by parents which somewhat explain their differences in attitudes towards pharmacological interventions. Firstly, parents noted

the difference between pharmacological interventions which were essential to cure cancer and pharmacological interventions which were not essential. Whilst they did not hesitate to administer essential pharmacological interventions to cure the disease, they preferred their child to tolerate symptoms or use non-pharmacological interventions to resolve symptoms. Secondly, parents noted the difference between milder pain which could be resolved with non-pharmacological interventions and severe pain which required pharmacological interventions:

*"I just feel that he's having so much medication, so I don't really want to keep giving him extra medication for pain and stuff if you could do it in another way... This medication he's having, he's got to have it whereas with pain medication, there's a choice, there might be something else that would work equally well, unless, if it's a high pain then obviously I would look at medical pain relief but otherwise I try and do something else." – Priya*

Importantly, many parents changed their attitudes towards pharmacological interventions throughout their child's cancer journey. Suzannah reported a stark change in her attitude towards morphine:

*"And there was one evening when Eleanor was particularly bad, and she was screaming... And my mother said well have you used that morphine? And I said, no I'm really scared of using the morphine. And she said, well she shouldn't be in this amount of pain... And I did. And it worked. And I realised I'd just been scared of giving her that for some reason. I think possibly because my grandmother died of cancer 10 years ago and she died at home and we were giving her morphine... But you know it's an amazing drug. Yes, let's have more of it." – Suzannah*

Finally, even if parents wanted to administer pharmacological interventions, at times they struggled to do this because children found them unpalatable:

*"...he wasn't gonna have it just for the taste of it. And then it put him off all the other ones that are actually okay to take" – Elena*

#### **6.4.4.4 Healthcare professionals**

When non-pharmacological and pharmacological interventions were either not preferred, or ineffective, the next intervention available was to call HCPs. Parents were aware of their community team, shared care centre, and principle treatment centre as potential sources of advice. Most parents expressed positive views of calling HCPs. Important aspects of calls to HCPs included availability, expertise, and relationship. Parents emphasised the importance of HCPs being constantly available:

*"...if I felt uneasy about his pain... I knew that I could talk to somebody 24 hours" – Pauline*

Not all HCPs were consistently available and individual availability influenced decisions of who to contact:

*“...whilst [nurse]’s our key worker at [our shared care centre], and [she’s] extraordinarily busy, she hardly spends any time at her desk, she’s not always easy to get hold of...So for me because it would be our [community] team” – Brenda*

In addition, parents stressed the importance of contacting someone with cancer expertise:

*“...the problem is when I call the [shared care hospital], unless it’s within working hours nine to five whatever, the oncology nurse isn’t there...because of his situation it makes more sense to call [principle treatment centre] so you’re talking to somebody who is au fait with the treatment” – Angie*

Parents emphasised the benefit of having a relationship with HCPs because every child is different:

*“...because...every child is different...what is absolutely crucial is having...a really good relationship with one or two people...who know your child, who are able to tell you exactly what you need to know” – Suzannah*

There was psychological benefit in knowledge that HCPs were available even if their services were not utilised:

*“Having contact numbers, knowing that people were at the end of the phone if I needed it...they’ve always said if you need anything, if it’s just a question, if she’s crying...just pick up the phone and just ring us. And because of that support...that’s taken us our anxiety down, knowing that they are there and we can pick up the phone.” – Stacey*

Parents also described limitations to this intervention. A few parents expressed dissatisfaction with information provided when they phoned HCPs. Brenda described a pain episode when she administered morphine for the first time. She knew the dose was insufficient because it was based on the lower weight of her child several months prior. She had called HCPs, but they had not permitted her to increase the dose:

*“I wish the doctor had...reviewed the dose he was, he had given and the fact that we could have had that as a baseline and then potentially given him more. But of course, she was dealing with a mother, not knowing me, not knowing Ollie, not knowing the full case history really” - Brenda*

Parents also described barriers to contacting HCPs such as not knowing when to call:

*“I didn’t know how severe things had to be before I contacted anybody...there’s a big cloud over this when you go home of actually who should you contact and why you can contact them.” – Jackie*

Several parents worried they would be perceived as “paranoid” if they called HCPs. Most parents were able to move beyond this as Ruth describes:

*“...you don’t want to be seen as this paranoid mother. But at the end of the day, god your child’s got cancer. So yeah, I think the first couple of times I phoned up I think I was a bit oooo should I be doing this? And then after that I thought, I don’t care, that’s what it’s there for. Just ring up. And I would say I’m sorry if it’s just a bit of a silly call but I’m gonna be a paranoid mum, can you tell me this.” – Ruth*

Lastly, parents hesitated to call HCPs because they predicted that call would inevitably lead to hospital attendance:

*“I know when I pick up the phone to my local that I’m going to drive in ...they can’t diagnose over the phone.” – Georgia*

#### **6.4.4.4.5 Hospital**

Parents’ final intervention, which they used as a last resort, was taking their child to hospital. Parents disliked this intervention due to the disruption it caused to the family unit and the high value they placed on time at home. This caused parents anxiety and they recalled uncertainty and worries about whether to go to hospital:

*“I didn’t know why I could go there and at which points I was supposed to go there and also I think you spend so much time in hospital you don’t want to go there for something silly cos you’re worried they’re gonna keep you in. That sounds silly cos on the other hand I don’t want her to be ill but I, if there’s nothing really wrong with her, then I want to spend the time that we’ve got at home at home. We sometimes used to sit there and...[my husband] would say, oh if you go to [hospital] you’ll be in there all week...everything goes through your head because you do as much as you can to stay out of being in hospital” – Jackie*

In the quote above Jackie worried about being kept in hospital unnecessarily but David expressed a divergent view:

*“...you could just come straight home if it turned out you didn’t need to go in. So, it’s less of a big deal.” – David*

Most parents recalled instances in which they had taken their child to hospital because of problematic pain which could not be resolved:

*“...we drove to the hospital...listening to her in the back of the car pleading with me...saying, please mummy. She was in such pain now looking back. I think at the time I sort of put the shutters down, we just need to get to hospital, seen by a doctor, all will be fine...It was really scary, and I think I did slightly block it all out and be like let’s just get on with the job of getting her to hospital” – Georgia*

Another reason for going to the hospital was being unsure of the cause of pain:

*“...if he was in genuine pain and it wasn’t something that I could easily explain would be to take him to the hospital” – Justin*

Other parents described going to hospital as a way of exercising caution:

*“I always err on the side of caution that I think oh well we ought to go and get him checked out” – Helen*

Sometimes it was the child who wanted to go to hospital:

*“I said, do you want to go to hospital? And she was like yes yes yes” – Stacey*

Sometimes pharmacological interventions had not relieved the child’s pain, so parents decided to go to hospital:



*“he had quite a bit of back pain and that did make him quite upset cos he was really, at that time we didn’t have any stronger...so I did take him to [hospital]...then he got stronger pain killers” – Pauline*

Despite the disruption it caused, and a desire to stay out of hospital, some parents described a sense of relief experienced in hospital:

*“It was a bit of a pain but then it’s peace of mind at the same time” – Helen*

*“...even though you’re in hospital, and sometimes that can feel...quite a relief. Cos you’re like, somebody else is going to look after her now.” – Georgia*

#### **6.4.4.4.6 Parent response: Pain assessment**

*“Like I said, every child is different and probably one more child with leukaemia will have a different, a different behaviour” – Natalia*

Parents’ response in the intervention phase was to assess their child’s pain. As Natalia describes above, the child’s uniqueness affected their pain expression behaviour and consequently pain assessment. Accurate pain assessment enabled parents to select an appropriate intervention. At times, pain assessment was easy as some parents reported their children telling them when they were in pain:

*“...when he’s in pain he lets me know, he will let me know” - Elena*

More frequently, parents reported their child would not articulate their pain:

*“And it’s a question of second guessing and working out what she needs cos she’s not that explicit about saying what works for her.” – Margret*

Parents occasionally admitted not knowing whether their child was in pain:

*“Well you don’t always, that’s the thing about feeling helpless. You don’t know but you just try and make a good guess really.” – Beth*

This uncertainty was in part caused by stoic children who appeared to have high pain thresholds and did not cry. Parents suggested reasons for this stoicism which differed with age. Older children often preferred not to talk about pain and wanted to cope without intervention. Pauline describes James choosing to “ride it” in the quote below:

*“I feel he does have pain but it’s not a pain enough to make him say, he will just ride it for a few hours...he will think...I’ll just wait it out and he’ll just ride the pain to a certain extent” – Pauline*

In the middle age group, parents felt their child was aware that if they admitted to being in pain, their parent would give them medication or take them to hospital. Children tried to hide their pain because they did not want these interventions:

*“...I think she was probably in more pain and...she didn’t want to talk. I think she didn’t want the medicine” – Jackie*

*“... not trusting that she was covering stuff up because she knew that if something hurt we were going to end up back in hospital...” – Georgia*

Parents of younger children felt unsure of their child’s pain due to their child’s inability to express pain:

*“I suppose obviously at her age, particularly a bit earlier on, she’s a bit older now but at the start of the process obviously her ability to communicate or articulate, in a way that an older child would be able to do is a challenge” – David*

Despite challenges, parents developed alternative ways of knowing when their child was in pain including attending to their child’s unique behavioural cues, mood, body language, verbal cues, and circumstances. Several parents reported that their child became quiet and did not talk or play, often withdrawing physically and trying to be alone:

*“We can tell because normally Raj’s quite a chatty person...we know he’s going through something...when he doesn’t want to talk to you” – Priya.*

*“She’ll go to be on her own...try and hide” – David*

For some children behavioural cues like being rude, moody, or angry were signs of pain:

*“He’ll snap at me go ‘what’ things like that which I know that’s not him” – Pauline*

*“She normally starts being moody and getting...then she’ll say oh my legs are really achy” – Lisa*

*“Throwing stuff like being angry with others...even though he needed me all the time he was very angry with me at the same time because it was...you was supposed to look after me” – Jana*

Body language was another way parents assessed their child’s pain. As with behavioural cues, body language was specific to the child. David described how his daughter would:

*“Collapse her shoulders a bit and droop” - David*

Suzannah described her daughter as:

*“Tense and clutching [her] stomach” – Suzannah*

Elena described a unique position which her child assumed when in pain:

*“...fold his leg over the other one kind of like a pretzel...was his little, his safeguard” – Elena*

Some parents observed their child extensively and learned to assess their child’s pain through creative attention to detail:

*“I...learned a lot from watching her play and...knowing how she was, how much she was hurting...if she was saying to the dolly oh you need this because you’re feeling like this...children can only go on what they’ve learned or what they know so if she’s saying that that dolly’s in pain cos this is how much it hurts and that’s what’s happened to her.” – Jackie*

In the excerpt below, Pauline describes how she could identify the intensity of her son's pain by noticing whether he was watching YouTube or using his PlayStation:

*"...he'll just watch YouTube he won't even go on the PlayStation cos I can tell when he's really wanting to rest and can't focus on the PlayStation game that will go and he'll lie down in bed and just watch, just watch YouTube or he'll put a video or a film on or something. And I will know then that he's still not quite well enough" – Pauline*

Differentiating between fear and pain was a further challenge to pain assessment as children in this sample experienced many fear-provoking circumstances:

*"...is it fear causing the pain? Is it anticipation of pain? Are these anxiety related symptoms?" – Suzannah*

Whilst parents generally felt their child's pain was genuine, a few parents, particularly of younger children, alluded to their child pretending to have pain to gain attention. These parents used circumstances to determine pain authenticity:

*"...she may be in trouble so she she'll say oh my finger when it's nothing to do with her finger" – Stacey*

#### **6.4.4.5 Consequence**

Depending on the success of the chosen intervention, the consequence would be one of two things: an "empty toolbox" of pain management interventions or a re-stocked pain management toolbox.

##### **6.4.4.5.1 Empty toolbox**

*"...but that sense of fear of feeling like the toolbox that you've grown up as a parent...suddenly all that's taken away from you. And you feel like you're back at square one...and in the most stressful situations." – Georgia*

If parents felt their intervention was unsuccessful, they were left feeling undermined and without any tools to effect change in their child's pain. They felt that their parenting skills had been removed and were aware that interventions available to them prior to diagnosis were no longer available. Parents felt there was nothing they could do:

*"I: what do you do if you think it is genuine sore tummy?"*

*P: Not a lot. Obviously, we can't really give her pain relief...I don't think there's much else we do." – David*

*"She says it doesn't work and nothing works...she would say pain was no better...I didn't have any other options." – Margret*

Often their "empty toolbox" was due to not feeling able to administer pharmacological interventions. Parents used several metaphors to describe the situation:

*“... [being able to give paracetamol is] almost like a safety blanket you know it’s there. But the moment you don’t have it you just think oh my god...what am I going to do?” – Stacey*

*“...they’ve taken it out of the bag of goodies” – Margret*

*“I went on a journey with the backpack only half packed. I needed a bit more. So that’s the way I felt about it.” – Suzannah*

#### **6.4.4.5.2 Re-stocking the toolbox**

Parents described increasing their repertoire of pain management techniques, through aspects of pain management already described such as knowing their child, non-pharmacological interventions, and pain assessment. Their re-stocking of the toolbox often involved creative interventions, flexible interventions and life adaptations:

*“Try to think outside the box...And trying alternatives...And just accept that sometimes you just have to go with the flow would be the bottom line.” – Margret*

*“...having a range of strategies and being flexible...so looking at each situation.” – Suzannah*

*“...at home, I think we’ve adapted quite well to even just little things...so we don’t cause pain...we’ve all adapted our behaviours...to minimise his pain” – Laura*

#### **6.4.4.5.3 Parent response: Fluctuating confidence**

Parents’ confidence fluctuated depending on the consequence of each intervention. Generally, parents began with low confidence regarding pain management. They repeatedly said they did not know what to do, what was happening in their child’s body, and how the treatment was affecting their child:

*“...he was screaming and things like that. And I...put him in a blanket but he didn’t stop so I was like oh my god maybe there’s something going on and I can’t help him but after 15 minutes, 20 minutes he stops...I’m not feel confident. I don’t know what’s going on with him.” – Natalia*

Pain episodes with a positive outcome increased parents’ confidence:

*“I’m more confident because many things happened so far, and I dealt with it in the right way so if we went through this we can [get] through other things as well.” – Jana*

Some parents reached a place of total confidence:

*I: How’s your confidence now?*

*P: Absolutely fine, absolutely fine. I don’t, everything is just second nature.”*

*– Stacey*

Some parents, whilst positive regarding their confidence, expressed caution due to a perception of not having experienced severe pain:

*“But the tumour itself hasn’t been painful or wasn’t painful which is lucky...So in general I feel confident, but I don’t think I’ve been fully tested, I don’t think she’s been high up the scale compared to some kids.” – David*

Some parents’ confidence had been on an upward trajectory but a pain episode with a negative consequence had lowered it again:

*“I did feel a lot more confident and lulled into what now turns out to be false sense of security. Because then when she did suddenly have this pain that I didn’t recognise, it completely threw me.” – Georgia*

This fluctuating confidence meant parents’ management of children’s cancer pain at home was a cyclical process. An increase in confidence at the consequence phase led to a decrease in pressure at the context phase and conversely a decrease in confidence at the consequence phase led to an increase in pressure at the context phase. Consequences of each cycle influenced the context of the next and the cycle repeated.

## **6.5 Summary**

This chapter provided results of three separate analyses of surveys, pain diaries, and interviews. In surveys, no participants were identified as displaying acquiescent response bias, or as having misunderstood or misread reverse items. Although both PPEP and MAQ total scales were distributed normally, only two sub-scales were normally distributed. Outliers were analysed but no evidence could be found to warrant removing these responses from the dataset. The proportion of parents choosing to participate did not statistically significantly differ from the proportion of parents choosing not to participate in terms of child age, child gender, or parent relationship to child. Descriptive statistics were used to present responses to each questionnaire statement and describe survey sample in terms of child and parent demographic characteristics. Inferential statistics have been presented. A statistically significant difference was found between parents’ age and MAQ, as well as parent ethnicity and PPEP, PPEP active loud, MAQ, and MAQ avoidance. Survey scores did not improve with time since diagnosis.

The proportion of parents choosing to extend their participation to complete a pain diary did not statistically significantly differ from the proportion of parents choosing only to complete a survey in terms of child age, child gender, or parent relationship to child. For most episodes, parents reported children were not in pain. Pain was most frequently reported in abdomen, legs, mouth/throat and head. Pain was most commonly caused by chemo-toxicity and constipation/diarrhoea. An increase in pain score was statistically significantly associated with parents responding with a combination of pharmacological and non-pharmacological interventions. In 207 (71%) episodes where children were in clinically significant pain, no analgesic drug was administered. Analgesic drugs were only administered in half of the episodes which were reported

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as having a pharmacological intervention response and paracetamol was parents' most frequent choice. Parents were more likely to respond with a non-pharmacological intervention than administer a pharmacological intervention. Most frequent non-pharmacological intervention responses were cuddles, food/drink, distraction, massage and sleep/rest. Parents reported an improvement in pain for 76% of episodes. Qualitative content analysis of free text responses revealed four categories and nine subcategories of reasons for no intervention.

Interview results suggested parents' management of children's cancer pain at home is a cyclical process with the key concept "every child is different" threaded throughout. In the context phase, children's pain manifestation, experience of other symptoms, and family context led parents to experience pressure. In the learning phase, parents used information, their personal background and informal support to become an expert in their child. In the intervention phase, there was a two-way process between parent assessment and five interventions: prevention; non-pharmacological interventions; pharmacological interventions; HCP; and hospital. In the consequence phase, parents either had an "empty toolbox" or re-stocked their "toolbox" which led to fluctuating confidence. This then added further context for the next cycle.

## Chapter 7. Integration of results

### 7.1 Overview of integration chapter

Chapter 6 provided details of the analysis of the individual datasets: surveys, pain diaries, and interviews. Within this chapter, the results of these datasets are integrated. Using a convergent parallel design (Creswell and Plano Clark, 2011), findings are integrated with the primary purpose of complementarity (Greene *et al.*, 1989), and the secondary purposes of completeness (Bryman, 2006), offset (Bryman, 2006; Petros, 2011), and explanation (Bryman, 2006). Integration embraces divergences and looks for new knowledge stemming from contradictions.

Integration is presented in three steps. Firstly, aggregate survey and pain diary results were integrated for the 37 participants who contributed to both of these datasets. This involved statistical investigations into data distribution and a comparison of scales and sub-scales to pain score, type of response, and pharmacological interventions. Secondly, integration was conducted by comparing aggregate survey, pain diary and interview results utilising matrices (Creswell and Plano Clark, 2011; Fetters *et al.*, 2013; Guetterman *et al.*, 2015). Inferences from each dataset were considered with reference to findings from the other datasets to distil inferences and facilitate step three. Thirdly, datasets were integrated using a joint display which focused on research questions (Creswell and Plano Clark, 2011; Fetters *et al.*, 2013; Guetterman *et al.*, 2015). Integration continues in Chapter 8 with a contiguous approach achieved through narrative. Figure 5.4 on page 85 displays this staged process pictorially.

### 7.2 Statistical integration of survey and pain diary data

#### 7.2.1 *Distribution of scales and sub-scales*

Appendix 12 shows results of the Shapiro-Wilk test to ascertain scale and sub-scales distribution. Both scales were normally distributed and parametric tests were used in their analysis. Sub-scales were a mixture of normal and non-normal distributions. As not all scales were normally distributed, tests involving both scales and sub-scales were analysed using non-parametric tests to ensure statistical assumptions were not violated.

#### 7.2.2 *Pain score compared to scales and sub-scales*

Table 7.1 provides comparisons of scales and sub-scales to mean, maximum, and clinically significant pain (pain score of three and above on NRS). A mild negative correlation was found

when the PPEP quiet-inactive sub-scale was compared to mean pain score and clinically significant pain. Having fewer misconceptions on the PPEP quiet-inactive sub-scale was mildly associated with rating both children's mean pain and clinically significant pain as higher.

### ***7.2.3 Type of response compared to scales and sub-scales***

Table 7.2 provides comparison of scales and sub-scales to parents' actions in response to pain. Parents' actions were grouped into four categories: administration of pharmacological interventions; administration of non-pharmacological interventions; a combination of pharmacological and non-pharmacological interventions; and no action. A moderate negative correlation was found when the PPEP quiet-inactive sub-scale was compared to the sum of episodes when a pharmacological intervention was administered to each child. Having fewer misconceptions on the PPEP quiet-inactive sub-scale was moderately associated with reporting more episodes in which pharmacological interventions were administered.

### ***7.2.4 Pharmacological interventions compared to scales and sub-scales***

Table 7.3 shows the results of pharmacological interventions compared to scales and sub-scales. A moderate negative correlation was found when comparing the PPEP quiet-inactive sub-scale to number of pharmacological interventions administered. A strong negative correlation was found when comparing the MAQ avoidance sub-scale to the number of times paracetamol was administered. Having fewer misconceptions on the PPEP quiet-inactive sub-scale was moderately associated with reporting more pharmacological intervention administrations. Having fewer misconceptions on the MAQ avoidance sub-scale was strongly associated with reporting more paracetamol administrations.



Table 7.1: Pain score in relation to scales and sub-scales

Independent variable	Dependent variable	Scale	Correlation coefficient	P-value
Maximum pain score	Scales <sup>†</sup>	PPEP	-.026	.881
		MAQ	.014	.939
	Sub-scales <sup>‡</sup>	PPEP active loud	-.024	.888
		PPEP quiet inactive	-.159	.347
		PPEP attention seeking	-.060	.727
		MAQ avoidance	-.058	.736
		MAQ appropriate use	-.050	.773
MAQ fear of side-effect	.037	.829		
Mean pain score	Scales <sup>†</sup>	PPEP	-.116	.333
		MAQ	-.103	.561
	Sub-scales <sup>‡</sup>	PPEP active loud	-.045	.790
		PPEP quiet inactive	-.356	.031
		PPEP attention seeking	.010	.954
		MAQ avoidance	-.019	.914
		MAQ appropriate use	-.162	.344
MAQ fear of side-effect	-.136	.430		
Clinically significant pain	Scales <sup>†</sup>	PPEP	-.090	.600
		MAQ	-.052	.771
	Sub-scales <sup>‡</sup>	PPEP active loud	-.054	.752
		PPEP quiet inactive	-.361	.028
		PPEP attention seeking	-.016	.924
		MAQ avoidance	-.34	.846
		MAQ appropriate use	-.105	.541
MAQ fear of side-effect	-.149	.385		

<sup>‡</sup>Spearman's correlation <sup>†</sup>Pearson's correlation

Table 7.2: Action in response to pain compared to scales and sub-scales

Independent variable	Dependent variable	Scale	Test statistic	P-value
Pharmacological intervention	Scales <sup>†</sup>	PPEP	r=-.071	.729
		MAQ	r=-.122	.570
	Sub-scales <sup>‡</sup>	PPEP active loud	rho=-.023	.912
		PPEP quiet inactive	rho=-.413	.036
		PPEP attention seeking	rho=.127	.538
		MAQ avoidance	rho=-.047	.825
		MAQ appropriate use	rho=-.063	.760
MAQ fear of side-effect	rho=-.127	.545		
Other (non-pharmacological)	Scales <sup>†</sup>	PPEP	r=.234	.197
		MAQ	r=-.062	.742
	Sub-scales <sup>‡</sup>	PPEP active loud	rho=.156	.374
		PPEP quiet inactive	rho=-.207	.257
		PPEP attention seeking	rho=.023	.899
		MAQ avoidance	rho=-.001	.996
		MAQ appropriate use	rho=-.129	.489
MAQ fear of side-effect	rho=-.240	.186		
No action	Scales <sup>†</sup>	PPEP	r=-.094	.739
		MAQ	r=.152	.588
	Sub-scales <sup>‡</sup>	PPEP active loud	rho=-.053	.851
		PPEP quiet inactive	rho=.174	.535
		PPEP attention seeking	rho=.258	.353
		MAQ avoidance	rho=.364	.182
		MAQ appropriate use	rho=.185	.510
MAQ fear of side-effect	rho=.350	.201		
Combination	Scales <sup>†</sup>	PPEP	r=.075	.783
		MAQ	r=-.080	.769
	Sub-scales <sup>‡</sup>	PPEP active loud	rho=.153	.571
		PPEP quiet inactive	rho=-.146	.588
		PPEP attention seeking	rho=.007	.980
		MAQ avoidance	rho=-.097	.722
		MAQ appropriate use	rho=-.140	.606
MAQ fear of side-effect	rho=.040	.883		

<sup>‡</sup>Spearman's correlation <sup>†</sup>Pearson's correlation

Table 7.3: Number of pharmacological intervention episodes compared to scales and sub-scales

Independent variable	Dependent variable	Scale	Significance	P-value
No. of pharmacological interventions administered	Scales <sup>†</sup>	PPEP	r=-.137	.050
		MAQ	r=-.086	.809
	Sub-scales <sup>‡</sup>	PPEP active loud	rho=.090	.663
		PPEP quiet inactive	rho=-.446	.022
		PPEP attention seeking	rho=.072	.726
		MAQ avoidance	rho=-.062	.768
		MAQ appropriate use	rho=-.096	.640
MAQ fear of side-effect	rho=-.154	.463		
No. of analgesic drug administrations	Scales <sup>†</sup>	PPEP	r=.006	.983
		MAQ	r=-.084	.912
	Sub-scales <sup>‡</sup>	PPEP active loud	rho=.114	.662
		PPEP quiet inactive	rho=-.203	.435
		PPEP attention seeking	rho=.031	.907
		MAQ avoidance	rho=-.388	.137
		MAQ appropriate use	rho=-.106	.684
MAQ fear of side-effect	rho=-.280	.277		
No. of paracetamol administrations	Scales <sup>†</sup>	PPEP	r=-.387	.214
		MAQ	r=-.375	.579
	Sub-scales <sup>‡</sup>	PPEP active loud	rho=.014	.965
		PPEP quiet inactive	rho=-.509	.091
		PPEP attention seeking	rho=-.161	.618
		MAQ avoidance	rho=-.702	.016
		MAQ appropriate use	rho=-.162	.614
MAQ fear of side-effect	rho=-.299	.345		
No. of morphine administrations	Scales <sup>†</sup>	PPEP	r=.420	.348
		MAQ	r=.446	.316
	Sub-scales <sup>‡</sup>	PPEP active loud	rho=.107	.819
		PPEP quiet inactive	rho=.273	.554
		PPEP attention seeking	rho=.436	.328
		MAQ avoidance	rho=.414	.355
		MAQ appropriate use	rho=-.036	.939
MAQ fear of side-effect	rho=.036	.939		

<sup>‡</sup>Spearman's correlation <sup>†</sup>Pearson's correlation

### 7.2.5 Outcome compared to scales and sub-scales

Free text comments in the outcome section of the pain diaries were not sufficiently specific to be able to be accurately categorised into “positive”, “negative” or “no change”. Quantitising of these data was subjective and it was not possible to meaningfully apply inferential statistics to these data. As a result, these data were analysed in the pain diary dataset (Section 6.3.8) but not integrated.

## 7.3 Integration using matrices for each data collection method

Meta-inference generation began as displayed in Figure 7.1 through integrating findings from each data collection method using matrices (Creswell and Plano Clark, 2011; Fetters *et al.*, 2013; Guetterman *et al.*, 2015). Three matrices were constructed for survey findings (Table 7.4), pain diary findings (Table 7.5), and interview findings (Table 7.6). Each finding was summarised in a written format and displayed in the first column. Subsequent columns were used to summarise the way in which each of the results of other data collection methods converged or diverged from each finding. Meta-inferences are displayed in the final column. Each matrix demonstrates part of the integration process and shows a step in how integration was achieved.

The matrix of survey findings (Table 7.4) led to an examination of reasons for divergence between datasets. Meta-inferences regarding MAQ suggested practical barriers may be the cause of negativity towards pharmacological interventions and that HCPs may be contributory to these negative attitudes. This matrix also suggested the meta-inference that the PPEP may not be able to detect all forms of pain expression due to every child with cancer being different.

Meta-inferences stemming from pain diary findings (Table 7.5) suggested that, due to convergences, pain diary and interview data collection methods measure the same phenomena. These meta-inferences confirm the most frequent location and causes of children’s pain. Although children were frequently reported as not being in pain, it was difficult to tell from the matrices whether this was due to parents being unable to assess pain. There was divergence between attitudes and behaviour around pain management: In interviews parents expressed a belief that they would administer pharmacological interventions if their child needed it but pain diaries revealed that as pain increased parents were not more likely to administer pharmacological interventions. Parents expressed a clear preference for non-pharmacological interventions and there was convergence on which pharmacological strategies parents prefer. Parents preferred paracetamol to other pharmacological interventions. Reasons for not administering pharmacological interventions were provided. Pain diaries and interviews converged on the finding that when children’s pain was rated as higher, this was associated with parents providing a wider variety of interventions.

Interview findings presented via the matrix in Table 7.6 revealed convergence between pain diaries and interviews which suggested they were measuring the same phenomenon. Once again meta-inferences regarding pain assessment were unclear. Divergences result either from parents not being able to assess their child's pain, or PPEP not being able to detect children's unique pain expressions. Preference for non-pharmacological interventions was confirmed as was the divergence between parents' attitudes and behaviour around pain management.

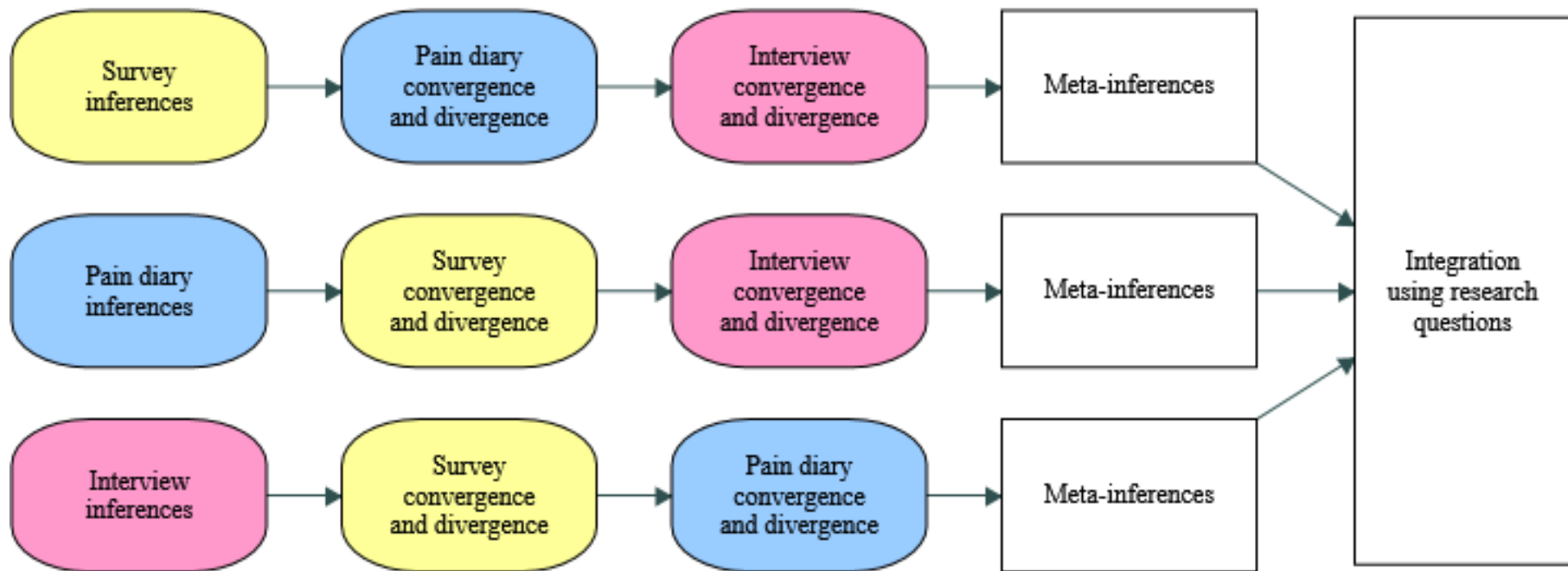


Figure 7.1: Integration of each data collection method using matrices

Table 7.4: Meta-inferences generated from survey findings

<b>Inference</b>	<b>Pain diary convergence</b>	<b>Pain diary divergence</b>	<b>Interview convergence</b>	<b>Interview divergence</b>	<b>Meta-inference</b>
Negativity towards analgesic drugs	As pain intensity increased, parents were not more likely to use pharmacological interventions. Non-pharmacological most frequent response. 71% of cases of clinically significant pain, no analgesic drug administered.	Some administration of pharmacological interventions evident. Several aspects of MAQ showed no statistically significant differences when compared to pain diary.	Negative attitudes explained: Child doesn't want analgesic drug; Constipation/nausea side-effects of morphine; Fear of masking a raised temperature; Stigma, experience of morphine in end of life situations; HCPs perceived as anti-analgesic drug.	Some very positive attitudes. Participants advised other parents to give pharmacological interventions.	Negative attitudes stemmed from practical barriers:  Pharmacological properties (e.g. masking temperatures, constipation); child refusing.  Interview sample have fewer negative attitudes than survey sample.  Survey is misleading to parents.
No improvements in pain medication attitudes over time over time	No significant differences in maximum or mean pain score with time since diagnosis.	None	HCPs were contributory to parents' negative attitudes and misconceptions. Many reasons for negativity did not change over time.	Parents reported changes in attitudes towards pharmacological interventions.	MAQ may not be sensitive to the nuances of pain management for children with cancer.  HCPs contribute to negative attitudes.  Interview sample have fewer negative attitudes than survey sample.

<b>Inference</b>	<b>Pain diary convergence</b>	<b>Pain diary divergence</b>	<b>Interview convergence</b>	<b>Interview divergence</b>	<b>Meta-inference</b>
Misconceptions about pain assessment	Children frequently not in pain. Parents who had better scores on PPEP quiet-inactive scale also reported their child experienced more pain and recorded more episodes of analgesic drug administration.	Several aspects of PPEP showed no statistically significant differences when compared to pain diary.	Few parents felt their child would occasionally fake pain.	Parents described in detail how they knew their child was in pain and put effort into understanding / assessing their child's pain.	"Every child is different" therefore PPEP may not be able to detect unique pain expressions.  Parents are mistaken in their belief they are able to assess pain.
No improvements in pain assessment over time	No significant differences in maximum or mean pain score time since diagnosis.	None	Stoicism, not wanting pharmacological interventions / hospital, age of child prevented parents knowing.	Parents described using behavioural cues, mood, body language, verbal cues, and circumstances to assess pain.	Interview sample are better at pain assessment than survey sample.  Survey is misleading to parents.



Table 7.5: Meta-inferences generated from pain diary findings

Inference	Survey convergence	Survey divergence	Interview convergence	Interview divergence	Meta-inference
Children frequently not in pain	None.	Sample endorsed many misconceptions on PPEP.	Parents described themselves as “lucky” due to the lack of pain their child had experienced.	Some parents commented that taking part in interviews made them realise their child was in regular low-level pain. Most parents could recall times when their child had severe pain at home.	Either children with cancer infrequently experience pain at home or parents have limited ability to detect pain.
Frequent pain locations	n/a	n/a	Similar locations.	Bottom higher emphasis in interview. Few mentions of headaches.	Pain diary and interview measure same phenomenon.  Children with cancer experience pain most frequently in abdomen, legs, mouth/throat, head, and bottom.
Cause of pain	n/a	n/a	Similar causes.	None	Pain diary and interview measure same phenomenon.  Most frequent causes of pain in children with cancer are: chemotherapy toxicity, constipation/diarrhoea, mucositis, and other drugs.
Parents use analgesic and non-analgesic in combination with high pain intensity	n/a	n/a	Evidence of parents using range of interventions and escalating actions as pain increased.	None	Parents widen repertoire of interventions in response to high intensity pain.

Inference	Survey convergence	Survey divergence	Interview convergence	Interview divergence	Meta-inference
As pain intensity increased, parents were not more likely to use analgesic interventions	Sample endorsed many misconceptions on MAQ.	None	Negative attitudes explained: “Not allowed” to give analgesic drug; Child doesn't want analgesic drug; Constipation/nausea side-effects of morphine; Fear of masking a raised temperature; Stigma, experience of morphine in end of life situations; HCPs perceived as anti-analgesic drug.	Parents said they would use pharmacological intervention if their child's pain increased. Some very positive attitudes in interviews. Parents advised other parents to give pharmacological intervention.	Parents believed they would administer pharmacological interventions if their child needed it, but evidence suggests this is not the case. There is a contradiction between attitude and behaviour.  Parents hold negative attitudes but do not admit to them / are unaware of them.  Interview sample hold fewer negative attitudes than survey sample.
No analgesic drug administered in 71% episodes of clinically significant pain					
Paracetamol most frequent drug administered	Better MAQ avoidance scores associated with more paracetamol administrations.	n/a	Negative attitudes towards morphine: Stigma, experience of morphine in end of life situations; Constipation/nausea side-effects of morphine; HCPs told parents better to be morphine naïve.	Negative attitudes towards paracetamol: “Not allowed” to give paracetamol; HCPs told parents not to give paracetamol; Fear of masking a raised temperature.	Despite pharmacological limitations of paracetamol, parents feel most comfortable administering it compared to other pharmacological interventions.
Non-pharmacological response most frequent response	Sample endorsed many misconceptions on MAQ.	None	Parents expressed preference for non-pharmacological response. Parents explained reasons for not giving pharmacological interventions.	None	Parents prefer non-pharmacological interventions.
Most frequent non-pharmacological responses	n/a	n/a	Similar non-pharmacological responses described in the survey.	None	Cuddles, food and drink, distraction, massage preferred non-pharmacological pain interventions.

<b>Inference</b>	<b>Survey convergence</b>	<b>Survey divergence</b>	<b>Interview convergence</b>	<b>Interview divergence</b>	<b>Meta-inference</b>
Reasons for non-pharmacological intervention	n/a	n/a	Reasons found in pain diary match closely to those described in the interview.	None	Analgesic administration limited by perception that pain is not severe or will be self-resolving, inadequacy of pharmacological interventions, HCP influence, and circumstance.

Table 7.6: Meta-inferences from interview findings

Inference	Survey convergence	Survey divergence	Pain diary convergence	Pain diary divergence	Meta-inference
Pain manifestation	n/a	n/a	Frequent pain locations and cause of pain match interview. Children frequently not in pain.	None	Pain diary and interview measure same phenomenon.
Know your child	None	Sample endorsed many misconceptions on PPEP.	n/a	n/a	"Every child is different" therefore PPEP may not be able to detect the uniqueness pain expressions necessary for pain assessment.  Some parents do not learn to assess their child's pain.  Interview sample are better at pain assessment than survey sample.  Survey is misleading to parents.
Non-pharmacological interventions	Sample endorsed many misconceptions on MAQ.	None	Non-pharmacological response most frequent response. Most frequent non-pharmacological responses match interview.	None	Parents prefer non-pharmacological interventions. Cuddles, food and drink, distraction, massage preferred non-pharmacological pain interventions.
Pharmacological interventions	Sample endorsed many misconceptions on MAQ.	None	As pain intensity increased, parents not more likely to use analgesic interventions. 71% of cases of clinically significant pain, no analgesic drug administered. Reasons for non-pharmacological intervention.	None	Parents believed they would administer pharmacological interventions if their child needed it, but evidence suggests this is not the case. There is a contradiction between attitude and behaviour.  Parents hold negative attitudes but do not admit to them / are unaware of them.  Interview sample hold fewer negative attitudes than survey sample.

Inference	Survey convergence	Survey divergence	Pain diary convergence	Pain diary divergence	Meta-inference
HCP	n/a	n/a	Occasional use of HCP resulted in hospital admission.	None	Pain diary and interview measure same phenomenon.
Hospital	n/a	n/a	Infrequent use of hospital confirms parents' use of it as a last resort.	None	Pain diary and interview measure same phenomenon.
Pain assessment	None	Sample endorsed many misconceptions on PPEP.	n/a	Children frequently not in pain.	<p>Either children with cancer infrequently experience pain at home or parents are unable to detect pain children's pain.</p> <p>Some parents do not learn to assess their child's pain.</p> <p>Interview sample are better at pain assessment than survey sample.</p> <p>Survey is misleading to parents.</p>

## 7.4 Integration using joint display of research questions

Integration using matrices for each data collection method was a key step, which facilitated the development of meta-inferences from three relatively large datasets. Following this, a third phase of integration was conducted using joint display structured around research questions.

Diagrammatic representation of this process is presented in Figure 7.2. Table 7.7 displays findings from each data collection method as they relate to each research question. Findings were extracted from datasets and compared for convergence or divergence which can be between-method or within-method. Potential explanations for divergence are provided in the final column.

Pain diary and interview data were convergent on pain locations and causes but had within-method divergence on pain prevalence. Chemotherapy toxicity was the most frequently cited reason for pain as measured by pain diaries, followed by constipation and diarrhoea, mucositis, and other treatment drugs. Interview data were qualitative and therefore cannot quantify the most frequent causes of pain, but similar causes were regularly mentioned in interviews so results from data collection techniques are considered convergent. The overall meta-inference stemming from this finding was that pain for children with cancer at home is primarily caused by treatments.

Abdominal pain, followed by leg, mouth/throat, head, and then bottom pain were found to be the most common locations for pain as measured by pain diaries. Although interview data cannot quantify most frequent pain locations, regular mentions of each of these sites throughout interviews with potentially more emphasis on bottom pain, suggests general convergence between datasets. Pain diaries and interviews had within-method divergence regarding the prevalence of pain: both datasets simultaneously found pain to be present and absent. Data from pain diaries and interviews suggest children with cancer are not often in pain at home. Conversely, in most of the interviews, parents described distressing episodes of pain and indicated that some children experienced clinically significant pain for long durations as evidenced in pain diaries. A potential explanation of this within-method divergence, is that there may be not be a single pain trajectory. Children's cancer pain at home may be best described by heterogeneous pain trajectories.

Survey and interview data diverged on how parents assess their child's pain at home. Surveys found that parents held negative attitudes and misconceptions towards children's pain expression which did not change with time suggesting parents are not able to adequately assess their child's pain. Conversely, interview results suggest parents can assess their child's pain. Due to the effort parents put into learning (Section 6.4.4.3), with the objective of knowing their child (Section 6.4.4.3.4), parents understand their child's unique pain expression and feel they are able to identify when their child is in pain (Section 6.4.4.4.6). There are four potential explanations for this divergence: two parent-based explanations, and two method-based explanations.

Pain diaries and interviews converge regarding what parents do to manage their child's pain at home. Pain diaries quantitatively found that when children are in pain at home, they frequently do not receive pharmacological interventions. Parents used non-pharmacological interventions more than pharmacological interventions with cuddles being the most frequent response, followed by food and drink, distraction, and massage. These techniques were frequently referenced by parents in interviews where parents reported a preference for non-pharmacological interventions. This convergence leads to the meta-inference that although parents frequently under-medicate their child's pain, they do not fail to respond. Instead, parents respond to children's pain at home using a variety of non-pharmacological interventions. As with pain manifestation, convergence between pain diaries and interviews suggests these data collection methods are measuring the same phenomenon.

Investigations into influences of parents' choice of interventions to manage children's pain revealed between-method divergence. Surveys identified parents as holding negative attitudes towards analgesic medications which did not improve with exposure to healthcare settings. Interviews identified some negative attitudes towards pharmacological interventions, but most parents had resolved these attitudes with time. Instead of attitudinal barriers towards pharmacological interventions, interviews identified practical barriers to pharmacological interventions. This is supported by evidence from pain diaries. There are several potential explanations for this divergence.

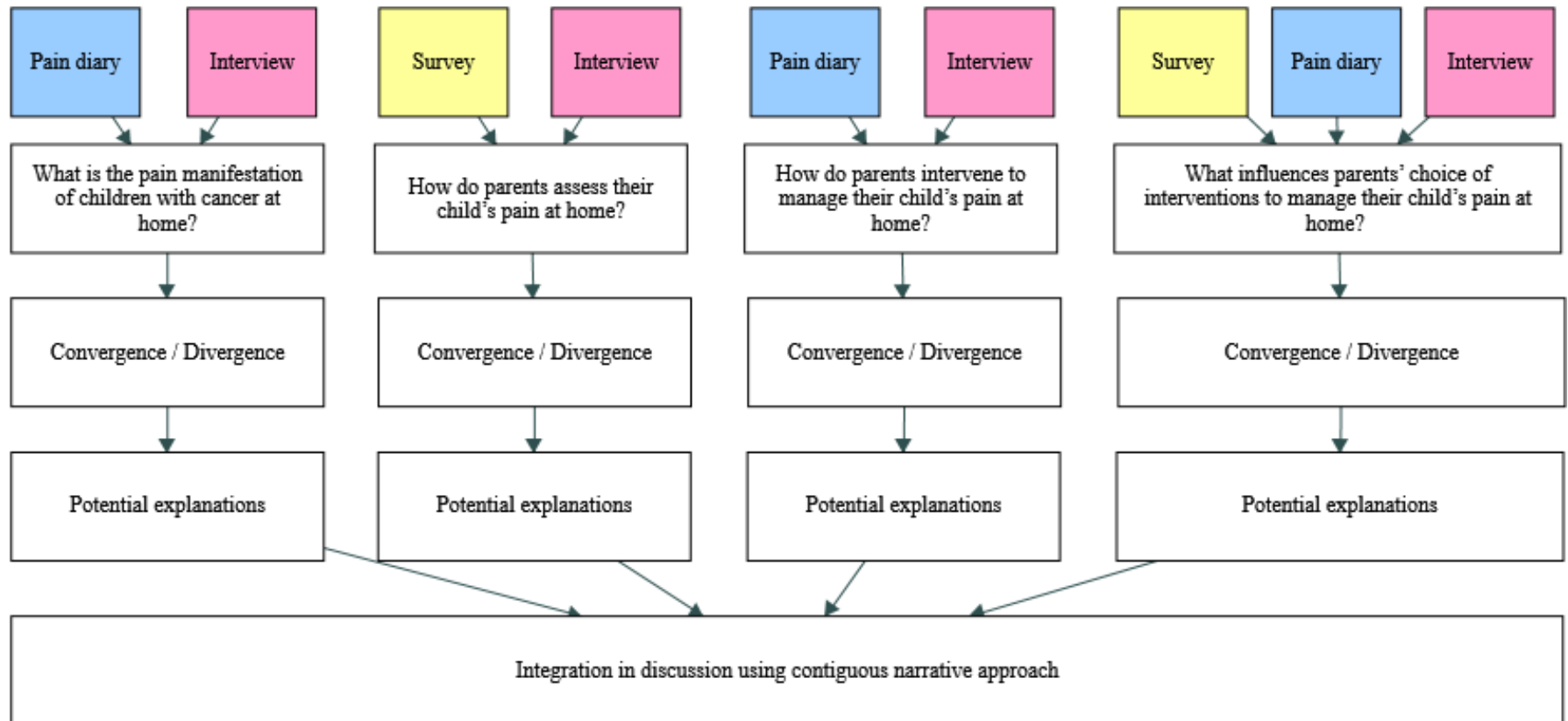


Figure 7.2: Diagrammatic representation of integration processes using joint display structured around research questions



Table 7.7: Joint display of integration structured around research questions

Survey	Pain diary	Interview	Meta-inference
<b>What is the pain manifestation of children with cancer at home?</b>			
n/a	<i>Cause</i>		
	Most frequent causes were chemotherapy toxicity, followed by constipation and diarrhoea, mucositis, and other treatment drugs.	Frequent mentions of chemotherapy toxicity, constipation, diarrhoea, mucositis, and other treatment drugs.	Convergence Pain in children with cancer is most frequently caused by treatments.
	<i>Location</i>		
	Most frequent locations were abdomen, followed by legs, mouth/throat, head, and bottom.	Frequent mentions of pain in abdomen, legs, mouth/throat, head and bottom.	Convergence Pain in children with cancer is most frequently in abdomen, legs, mouth/throat, head and bottom.
	<i>Prevalence</i>		
Children had no pain in 74% of episodes.	Parents reported being "lucky" with lack of pain, low level of pain, fleeting pain.	Within-method divergence Potential explanation: - Heterogeneous pain trajectories	
Clinically significant pain occurred at least once for 85% of children.	Parents described episodes of pain at home which could not be resolved.		

<b>How do parents assess their child's pain at home?</b>			
Parents hold many negative misconceptions regarding pain assessment which do not improve over time.	n/a	Each child has a unique pain expression. Parents know their child and use many techniques to make accurate assessment of their child's pain.	Between-method divergence Potential explanations: - Parents are mistaken in their belief they are able to assess pain. - Interview sample are better at pain assessment than survey sample. - Survey is misleading to parents. - Survey is insensitive to individual child pain expression.
<b>How do parents intervene to manage their child's pain at home?</b>			
n/a	Non-pharmacological response most frequent response. 71% of episode of clinically significant pain, no analgesic received. Most frequent non-pharmacological interventions were cuddles, followed by food and drink, distraction, and massage	Parents preferred non-pharmacological interventions.  Frequent references to distraction, cuddles, massage, food and drink.	Convergence Parents frequently under-medicate their child's pain but use many non-pharmacological interventions to manage it.

<b>What influences parents' choice of interventions to manage their child's pain at home?</b>			
<p>Parents hold many negative misconceptions regarding analgesic drugs which do not improve over time.</p>	<p>Practical barriers to pharmacological interventions noted in reasons for no action:</p> <ul style="list-style-type: none"> <li>- Child refuses pharmacological interventions.</li> <li>- Side-effects of analgesic drugs.</li> <li>- HCP advises against analgesic drug.</li> <li>- Not allowed to give analgesic drug.</li> <li>- Analgesic drug will not help.</li> </ul>	<p>Few attitudinal barriers detected, most resolved with time.</p> <p>Practical barriers to pharmacological interventions:</p> <ul style="list-style-type: none"> <li>- Paracetamol could mask a raised temperature.</li> <li>- Morphine could cause or exacerbate nausea or constipation.</li> <li>- Ibuprofen and codeine contraindicated.</li> <li>- Children refuse pharmacological interventions.</li> </ul>	<p>Between-method divergence</p> <p>Potential explanations:</p> <ul style="list-style-type: none"> <li>- Interview sample hold fewer negative attitudes than survey sample.</li> <li>- Parents hold negative attitudes but do not admit to them / are unaware of them.</li> <li>- Survey is detecting practical barriers (analgesic context and unpalatability of medication) not attitudinal barriers.</li> </ul>

## 7.5 Summary

This chapter integrated findings of this research in three steps. In the first step, quantitative data from the survey and pain diaries were statistically integrated. Integration of survey and pain diary data revealed parents who rated their child's pain as higher also had fewer misconceptions as rated by the quiet-inactive sub-scale. Parents who reported more episodes in which pharmacological interventions had been administered also had fewer misconceptions as rated by the quiet-inactive sub-scale. Parents who recorded administering more pharmacological interventions also had fewer misconceptions as rated by the quiet-inactive sub-scale. Parents who recorded more episodes of analgesic drug administration had fewer misconceptions as rated by the MAQ side-effects sub-scale. Parents who recorded more episodes of paracetamol administration had fewer misconceptions as rated by the MAQ side-effects and MAQ avoidance sub-scales.

Secondly, integration of survey, pain diary, and interview data were presented using matrices (Creswell and Plano Clark, 2011; Fetters *et al.*, 2013; Guetterman *et al.*, 2015) to sort three large datasets in a series of tables. In each table, inferences were presented alongside convergent and divergent evidence and finally meta-inferences were generated. This step facilitated the third level of integration, in which joint display was used to integrate data around research questions. Convergent and divergent integration resulted in potential explanations which will provide the basis for continued integration using a contiguous approach achieved through narrative discussion in the next chapter. Results of this integration are discussed in Chapter 8 with reference to other research and theoretical frameworks.

## Chapter 8. Discussion

### 8.1 Overview of discussion chapter

In this chapter, integration continues using a contiguous approach achieved through narrative (Fetters *et al.*, 2013). Introductory sections describe the relationship between research questions and summarises research findings. Following this, meta-inferences and potential explanations for convergences and divergences outlined in Table 7.7 are used as the basis for four sections describing findings of each research question in relation to the wider research literature. Discussion then returns to the findings of the integrative review in Chapter 3 before an in-depth analysis of findings in relation to theoretical frameworks. Final sections include discussion of strengths and limitations.

#### 8.1.1 Relationship between research questions

Understanding the relationship between research questions (Table 2.4) enhances understanding of the integration of the datasets within this research. Figure 8.1, shows how the research questions relate to each other and the Theory of Planned Behaviour. The research questions are interrelated and understanding of one research question affects understanding of the other research questions either directly or indirectly. This has two implications for the interpretation of findings. Firstly, awareness of findings from one research question will aid understanding when interpreting findings from another research questions. Secondly, it is important to be aware that findings from one research question are of limited use without findings of the other research questions.

In this research, due to the way in which children's pain manifestation was measured (by proxy using parent report), parents' assessment of their child's pain and pain manifestation are not mutually exclusive. Findings regarding children's pain manifestation both leads to and from parents' assessment of pain. In Figure 8.1, the two-way arrow from pain manifestation to pain assessment represents how children's pain manifestation influences parents' assessment of their child's pain. In addition, this arrow indicates the way in which, due to lack of a self-report measure, pain manifestation measured in this study is a product of parents' assessment and may not be purely based on the child's experience of pain. To evaluate the use of an intervention on an individual, it is necessary to know the extent of their pain so if children's pain manifestation is unknown, an understanding of interventions to manage that pain is of limited use. Parents' response to children's pain is similarly undermined if there is no understanding of how parents are assessing children's pain. Understanding of what influences parent responses to their child's pain cannot be ascertained without knowledge of what parents do to respond. The way in which parents respond to

their child's pain is a result of their child's pain manifestation (mitigated by pain assessment), pain assessment, and influences of parent response. Pain assessment and influences of parent response are moderated by TPB but due to their interconnected nature, TPB will indirectly relate to all research questions.

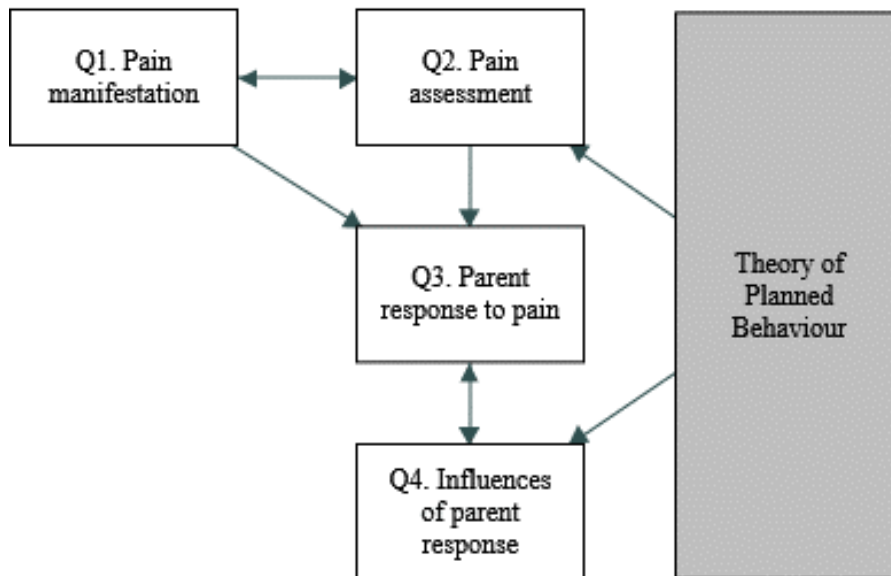


Figure 8.1: Relationship between research questions

### ***8.1.2 Overview of research findings***

A summary of research findings in relation to each research question is presented in Table 8.1. The following four sections discuss each research question applying literature critically. Discussion embraces divergences between and within research methods and seeks to identify the likelihood of each potential explanation suggested in Table 7.7 (page 160) being the true cause of divergence.

Table 8.1: Overview of research findings

<b>1. What is the pain manifestation of children with cancer at home?</b>	
<i>Convergence</i>	
Pain was primarily caused by treatment.	
Most frequent pain locations were the abdomen, followed by legs, mouth/throat, head, and bottom.	
<i>Divergence</i>	
Most of the time, children were not in pain at home.	Most children had episodes of clinically significant pain at home.
<b>2. How do parents assess their child's pain at home?</b>	
<i>Divergence</i>	
Parents put effort into learning their child and are able to assess their child's pain. Every child had a unique pain expression.	When measured quantitatively, parents held many negative misconceptions regarding pain assessment which did not improve over time.
<b>3. How do parents intervene to manage their child's pain at home?</b>	
<i>Convergence</i>	
Parents frequently under-medicated their child's pain.	
Parents preferred non-pharmacological interventions and used a wide range.	
<b>4. What influences parents' choice of interventions to manage their child's pain at home?</b>	
<i>Divergence</i>	
Parents held positive attitudes towards pharmacological interventions, but practical barriers left parents with an "empty toolbox" of pharmacological interventions.	When measured quantitatively, parents held many negative misconceptions regarding analgesic drugs which did not improve over time.

## 8.2 What is the pain manifestation of children with cancer at home?

Parents' perspectives of pain manifestation were recorded in the pain diaries and described in the interviews. Datasets converge on causes and location of pain but there was within-method divergence on prevalence of pain in children with cancer at home.

### 8.2.1 Causes of pain in children with cancer at home

Previous studies aiming to investigate causes of cancer pain in children at home, reported that participants may have misunderstood the purpose of data collection as they reported pain from procedures which could not have happened at home (Bossert *et al.*, 1996; Gedaly-Duff *et al.*, 2006). Although in the pain diaries parents occasionally reported procedures as a cause of pain at home, in interviews parents explained that whilst these procedures, for example lumbar punctures,

took place in the hospital setting, pain resulting from these procedures continued into home settings. There were no instances of non-cancer pain described in pain diaries suggesting participants had understood the purpose of data collection. Of note, in 5% of episodes, parents recorded not knowing the cause of the pain, and for four children (10%), this was the most commonly recorded cause. An unknown source of pain is a unique finding to research in this thesis and may be due to data collection procedures used. Previous research has not provided parents opportunity to state their uncertainty (Bossert *et al.*, 1996; Madi and Clinton, 2018). Evidence that parents had freedom to record pain cause, combined with evidence that parents understood the purpose of data collection, suggest these data are a valid representation of what parents in this sample believed had caused pain in children with cancer at home.

This research found treatment to be the primary cause of pain experienced by children with cancer at home. Conversely, a recent study in Lebanon of inpatient and outpatient children with cancer, found tumour and metastases were the most frequent causes of pain (Madi and Clinton, 2018). In that study no significant differences were found in pain intensity between pain caused by cancer, treatment, and procedures. Children were asked to choose between a predetermined list of potential causes of pain and authors suggested this may have biased or limited children's responses. Treatment as the major source of cancer pain in children is otherwise widely corroborated (Hanmod and Gera, 2016; Ljungman *et al.*, 2006). Findings reported in this thesis are in line with a historical trend toward more pain from treatment and less pain from the disease itself (Twycross *et al.*, 2015b).

### ***8.2.2 Location of pain in children with cancer at home***

The research presented in this thesis found the most frequent location of pain for children with cancer at home to be the abdomen, legs, mouth/throat, head, and bottom. Comparison with other literature is hindered by different ways of reporting pain location. In a study reporting on an app designed for managing cancer pain at home, a small sample (n=12) of outpatient children made no mention of abdominal pain when using a body diagram (Fortier *et al.*, 2016). In concordance with findings in this thesis, both abdomen and legs were within the top five pain locations in a sample of outpatient children though the mechanism of recording is unknown (Fortier *et al.*, 2014). A sample of inpatient and outpatient children similarly reported abdomen and head to be in the top five pain locations using a body diagram (Madi and Clinton, 2018). When given four options to choose from, parents of inpatient and outpatient children with cancer both on and off treatment, reported legs to be the most frequent location of pain (Tutelman *et al.*, 2018).



Pain location can help identify the cause of pain and subsequently its treatment. Leg pain is likely to be caused by peripheral neuropathy (Armstrong *et al.*, 2005; Wickham, 2007) which will need management with anticonvulsants such as gabapentin and pregabalin (Hanmod and Gera, 2016). Some abdominal pain may be linked to vomiting and if so, will require antiemetics as well as analgesic drugs (Hanmod and Gera, 2016). Mouth or throat pain could be caused by peripheral neuropathy requiring anticonvulsants, or resultant from mucositis, in which case oral hygiene measures may be required in addition to analgesic drugs (Miller *et al.*, 2012). Bottom pain and abdominal pain may be resultant from constipation which would require laxatives and dietary changes as well as analgesic drugs (Pashankar *et al.*, 2011; The Nemours Foundation, 2018). Awareness of the location of pain experienced by children at home will help HCPs equip parents to intervene and manage it effectively.

### **8.2.3 Prevalence of pain in children with cancer at home**

Pain diaries and interviews both had within-method divergence regarding the prevalence of pain: each dataset simultaneously found pain to be present and absent. Many pain diaries and interviews suggest children with cancer are not in pain at home very often. The absence of pain as a finding of this research suggests one of two scenarios: either children with cancer do not experience pain at home very often, or parents are limited in their ability to detect their child's pain. Although this research did not use self-report measures of pain making difficult to ascertain which scenario is true, previous research using self-report of pain intensity concluded that children generally do not experience severe pain at home (Fortier *et al.*, 2014). Combining these results, it can tentatively be suggested that the first scenario is more likely: children with cancer do not experience pain at home very often. Greater confidence in this explanation stems from an analysis of how parents assess their child's pain at home (Section 8.3.1).

Conversely, in most of the interviews, parents described distressing episodes of pain and in the pain diaries parents reported that some children experienced clinically significant pain for long durations. Despite what appears to be within-method divergence, these divergent findings may not be mutually exclusive. It may be possible for both to be true. A potential explanation for this is that there may be not be a single pain trajectory: children's cancer pain at home may be best described by heterogeneous pain trajectories. For example, there may be a subset of children who experience no pain at home, a subset of children who experience frequent pain at home, and a subset of children who experience occasional pain at home. This is corroborated by the key finding from interviews that "every child is different".

Heterogeneous pain trajectories could explain divergence of pain prevalence found in literature and why obtaining pain prevalence data for this population is challenging (Twycross *et al.*, 2015b). In a two-week daily diary study in children with cancer at home, overall pain severity was mild and parents frequently reported not administering pharmacological interventions due to their child not being in pain (Fortier *et al.*, 2014). In an online survey, parents of children with cancer were asked to estimate their child's average, worst, and least pain over the past month from 0-10 on the NRS (Tutelman *et al.*, 2018). Mean pain for children on active treatment was 3.53 (average), 6.34 (worst), and 0.97 (least) although it is unclear how much of this was pain at home. These combined data suggests that for children who do have pain, it is frequently clinically significant and at worst severe.

Due to data quality, statistical confirmation of heterogeneous pain trajectories could not be performed using pain diary data so evidence from literature will be used to support this potential explanation. A longitudinal study of children with cancer, provided evidence of two heterogeneous symptom trajectories which included pain: less severe and more severe (Wang *et al.*, 2017). Another study in children with cancer found evidence of four symptom and function profiles and related this to children having another medical condition (Buckner *et al.*, 2014). Both studies predicted each child's symptom trajectory using criteria which are frequently unavailable in clinical practice and authors of these studies emphasised difficulties in predicting profiles based on demographic criteria. This suggests HCPs in clinical practice may be unable to detect which children will experience more pain at home. An educational checklist has recently been developed which includes a list of topics for nurses to address with parents of children newly diagnosed with cancer prior to initial discharge (Rodgers *et al.*, 2018). On this list, pain and pain management are considered primary topics which only require attention if applicable. Research described in this thesis found that when children did experience pain, it was often clinically significant. Clinically significant pain episodes occurred at least once for 84% of children which implies this topic should be addressed in the vast majority of cases and it may not be appropriate to address this topic only as required. As it is currently not possible to predict which children will experience pain and when (Buckner *et al.*, 2014; Wang *et al.*, 2017), pain education prior to discharge must be a universal consideration for parents of all children with cancer regardless of their pain experience in clinical settings.

### **8.3 How do parents assess their child's pain at home?**

Data integration revealed divergences between datasets regarding how parents assess their child's pain at home with survey suggesting parents are not able to assess their child's pain at home and

interviews indicating the reverse. Potential parent-based and method-based explanations for this divergence are now considered.

### **8.3.1 Parent-based explanations for divergent findings**

Two possible parent-based explanations will be discussed. Firstly, parents may be mistaken in their perception that they are able to assess their child's pain. It is not possible to say for certain whether parents' pain assessments are accurate, as this research did not include a self-report measure of pain by children. Many studies reveal concordance between parent and child reports of pain. In qualitative work, child and parent perceptions of cancer pain have been found to be compatible (Ljungman *et al.*, 2006). A congruent relationship between parent and child reports of cancer pain was found in studies relating to procedure pain (Badr *et al.*, 2006), and longitudinal symptom assessment (Baggott *et al.*, 2012). Conversely, literature reports divergence between parent and child reports of pain. A meta-analysis which investigated dyadic concordance for all types of childhood pain concluded that despite moderate effect sizes, parent reports should only be considered an estimate of their child's pain (Zhou *et al.*, 2008). This meta-analysis included 12 studies, of which four investigated postoperative pain, and five procedural pain. Both of these pain manifestations are short-term, and parents may be less used to assessing their child's pain in these circumstances compared to parents of children with cancer. In children with cancer, one study found parents' retrospective recall of pain, amongst other symptoms, was higher than children's recall which is contrary to suggestions of under recognition of pain in this population (Zhukovsky *et al.*, 2015). This combined evidence suggests parent and child reports of cancer pain are congruent and parents are able to assess their child's pain.

An examination of the accuracy of parents' pain assessment can be made by considering the trustworthiness of proxy reports of children's pain using wider literature and the content of interviews. Assessment of the trustworthiness of proxy report of children's pain is made on the assumption that self-report is the gold-standard in pain assessment (Baggott *et al.*, 2014; Finley *et al.*, 2009). When a measure is considered gold-standard, this suggests it is accurate and should be unquestioned, but there are many reasons why clinicians and researchers may question the use of self-report as gold-standard (Twycross *et al.*, 2015c; Versloot *et al.*, 2013). Evidence suggests children give different self-reports of pain to different people (Versloot *et al.*, 2004, 2013). One explanation is the concept of "display rules", where a child is likely to display a behaviour (i.e. pain expression) if they perceive it will lead to a positive outcome and not if they perceive it will lead to a negative outcome (Versloot *et al.*, 2013). Display rules are evidenced in the research described in this thesis. Parents described children not wanting to admit to their pain due to fear of having to take pharmacological interventions which they found unpalatable, thinking pain would mean they

would have to go to hospital, and simply not wanting to think about pain (Section 6.4.4.4.6). It is, therefore, possible that any self-report measures of child pain utilised in this research may have been less accurate than parents' proxy report.

Current academic thinking suggests self-report should be considered the primary, but not the sole source of information for pain assessment (Twycross *et al.*, 2015c). Instead, it is suggested that approaches in which several aspects of child pain are simultaneously considered and weighed against one another may be more beneficial. These methods are termed "bundled approaches". One example of a bundled approach is the CARES (Context, Assessment, Risk, Emotion, Socio-cultural) approach which considers: context, assessment of pain using self-report and behavioural signs, risk factors, emotional factors including developmental and psychological considerations, and socio-cultural factors such as patient preferences. This is considered a bundled approach because as with other care bundles, it uses several different strands of information to make an assessment and deliver an intervention.

The intervention phase of the model set out in Section 6.4.4.4 describes how parents assessed their child's pain using a similarly bundled approach which closely matches CARES. Parents reported using circumstances surrounding the pain episode, children telling them when they were in pain, behavioural cues, considering emotions by differentiating between fear and pain, and considering their child's age and developmental stage. Parents balanced risks of analgesic drug administration and non-pharmacological interventions and used their own background and family preferences in managing pain (Section 6.4.4.3.2). Parents reported combining several elements of information in their assessment of their child's pain which constitutes using a bundle. In short, pain assessment as conducted by parents of children with cancer closely matches pain assessment as advised by current world experts in paediatric pain (Twycross *et al.*, 2015c). Although the research described in this thesis is the first to confirm the use of bundled approaches by parents of children with cancer, evidence of parents' use of aspects of the CARES bundled approach has been found in other children's cancer literature. Use of behavioural cues for pain assessment has been widely reported by parents of children with cancer (Bettle, 2015; Forgeron *et al.*, 2006; Irvine, 2017), with withdrawal and introversion by parents as signs of cancer pain in children (Lu *et al.*, 2011). Overall, this evidence suggests that whilst parents' pain assessment should be considered only an estimate of pain, it can be considered a good estimate. It is unlikely parents are mistaken in their understanding of their ability to assess their child's pain. The research presented in this thesis identified bundled approaches as an accurate way of measuring pain in children with cancer which researchers may find useful when designing future research and HCPs may find useful when assessing children's pain in clinical settings.

A second parent-based explanation is that, on average, interview participants may have been better at pain assessment compared to those completing the survey. Participants' prior pain assessment knowledge or education received from HCPs was unknown. Using a sampling frame ensured a range of participants in terms of age of child and time since diagnosis but it did not attempt to include parents with a range of pain assessment abilities. A recruitment strategy which ensured a range of participants in terms of pain assessment abilities would have been difficult to design and may have biased data collection if the researcher was aware of participants' pain assessment abilities when conducting interviews. Although a balanced interview sample is not guaranteed, inspection of the PPEP scores of interview participants (Table 6.14) revealed a range of pain assessment abilities which indicates that biased sampling may not be the cause of divergence between datasets. In conclusion, neither parent-based explanations appear to be the cause of divergence between datasets in this context.

### **8.3.2 Method-based explanations for divergent findings**

Two possible method-based explanations exist. Firstly, closer examination of the PPEP, suggests some questions may be misleading for parents. For example, parents who agree with the statement "children in pain have trouble sleeping" will be scored as having a misconception. Whilst being asleep does not mean that a child has no pain, children with pain resultant from a variety of causes do have trouble sleeping (Fortier *et al.*, 2014; Haraldstad *et al.*, 2011; Lynch *et al.*, 2018; Palermo *et al.*, 2011, 2012). Similarly, parents who agree with the statement "children who are playing are not in pain" will be scored as having a misconception. Although playing does not mean the child has no pain, problems with social functioning are associated with acute (Roth-Isigkeit *et al.*, 2005) and chronic pain (Fortier *et al.*, 2014; Palermo, 2000) in children so it may be possible that parents had misunderstood the question. It may not be appropriate to classify parents who agree with statements such as these as having a misconception in the context of children's cancer pain.

Secondly, the PPEP may not be sufficiently nuanced to detect the uniqueness of each child's pain expression. Interview results were focused around the key theme "every child is different", which describes how each child's uniqueness impacts parents' pain management. Due to the chronic nature of cancer pain in children (Fortier *et al.*, 2014), parents have time to learn their child's unique pain expression. Quantitative, closed-question data collection methods, such as surveys, may lack the sensitivity required to measure nuances of pain assessment in this population of children with a chronic condition. Conversely, open-ended data collection methods, such as interviews, allow parents to describe their child's unique pain expression and their unique method of pain assessment. Whilst there is evidence of PPEP reliability from acute settings (Zisk *et al.*,

2007a, 2010), its validity in measuring pain assessment in a context where parents have opportunity to learn their child's unique pain expression is unknown. This may explain why parents in this study seem to show more misconceptions on PPEP than non-cancer populations where the analgesic context is different (Twycross *et al.*, 2015d; Zisk *et al.*, 2007a, 2010) (Appendix 13).

In summary, divergence between datasets on pain assessment can be understood by examining parent-based and method-based explanations. Lack of self-report may mean the true nature of children's cancer pain at home is not fully represented. Evidence of child disclosure of pain being limited by display rules suggests self-report may not represent an unquestionable gold-standard in this context. Parents' use of several aspects of child pain expression is in line with expert recommendations to use bundled approaches to pain assessment. This evidence suggests parents are not mistaken in their understanding of their ability to assess pain. Participants who chose to complete interviews may be more attuned to their child's pain leading to a biased sample but inspection of interview participants' PPEP scores suggests this is not the case. Some PPEP questions may have misled parents and exaggerated misconceptions in pain assessment. Finally, because "every child is different", the PPEP may be insensitive to children's unique pain expression. Integration suggests method-based explanations are more likely the cause of divergence than parent-based explanations. Not only are parents able to adequately assess their child's pain but use of bundled approaches means they are in line with current expert thinking on children's pain assessment. Although parents experience challenges in pain assessment they overcome these challenges and can confidently assess their child's pain.

## **8.4 How do parents intervene to manage their child's pain at home?**

Parents recorded how they intervened to manage their child's pain in pain diaries and described this in interviews. Datasets converge on what parents choose to do to manage their child's pain at home.

### ***8.4.1 Parents under-medicated their child's pain and preferred non-pharmacological interventions***

Integrated results revealed that parents frequently under-medicated their child's pain. Confirmatory evidence of under-medication of children's cancer pain can be found in a two-week pain diary study which discovered that, on average, 2.5 children each day did not receive analgesic drug interventions despite having clinically significant pain (Fortier *et al.*, 2014). Fortier and colleagues did not attempt to measure administration of non-pharmacological interventions so children who did not receive an analgesic drug were classified as not having had their pain treated. An important

contribution of the research presented in this thesis is the knowledge that parents use a variety of non-pharmacological interventions to manage their child's cancer pain at home.

Integrated results revealed that parents prefer non-pharmacological interventions and use a wide range of these to manage their child's pain at home. Literature in support of non-pharmacological interventions to manage pain in children with cancer is growing (Jibb *et al.*, 2015). Children with cancer use more non-pharmacological strategies than other populations (Post-White *et al.*, 2009a). Parents use more non-pharmacological strategies to manage their child's cancer pain than pharmacological strategies (Tutelman *et al.*, 2018). Distraction is frequently the most common strategy used by parents to manage cancer pain (Bettle, 2015; Fortier *et al.*, 2016; Tutelman *et al.*, 2018). In interviews, parents of children with acute lymphoblastic leukaemia on treatment or 12 months post-treatment focused on non-pharmacological strategies which they considered to be the first step in pain management (Bettle, 2015). Parents may be under-medicating their child's pain but contrary to previous literature (Fortier *et al.*, 2014), research in this thesis concludes that parents are responding to their child's pain using a variety of non-pharmacological strategies. Parents' frequent use of non-pharmacological interventions shows that previous research which did not consider these interventions may have underrepresented parents pain management abilities.

## **8.5 What influences parents' choice of interventions to manage their child's pain at home?**

Having established how parents intervene to manage their child's pain at home, influences on parents' choices of intervention will be considered next. Research in this thesis found divergences between datasets regarding what influences parents' choice of interventions to manage their child's pain at home. Surveys found parents hold negative attitudes towards pharmacological interventions. In interviews, parents held positive attitudes towards pharmacological interventions but described practical barriers which are also documented in pain diaries. Parent-based and method-based explanations for this divergence will be discussed.

### ***8.5.1 Parent based explanations for divergent findings***

As with parental pain assessment, a potential explanation of this divergence is that the interview sample may have held fewer negative attitudes compared to the survey sample. Arguments to refute this potential explanation mirror arguments surrounding pain assessment attitudes which have been outlined previously in Section 8.3.1 and will not be reiterated here. Inspection of MAQ scores of participants included in interviews (Table 6.14) revealed a range of attitudes toward analgesic medications represented within the sample which suggests biased sampling may not be

the cause of divergence between datasets. In addition, pain diaries converge with interviews on influences of choice on interventions adding further weight to the argument that a biased interview sample is not the cause of between-method divergence (Table 7.7). Pain diary findings are able to add weight to interview findings due to the larger sample of parents included in pain diaries which further reduces the chance that the sample was biased when compared to surveys.

A further potential explanation is that parents may hold negative attitudes which they are either unwilling to admit to, or are unaware of, and are therefore mistaken in describing positive attitudes towards pharmacological interventions. Whilst it is not possible to state with certainty, several indicators suggest this is not the case. Firstly, in interviews, many participants reported improvement in attitudes towards morphine throughout the duration of their child's cancer treatment. Parents reported that they initially were uncertain about administering morphine due to previously held stigma, but once they saw the pain relief it could provide to their child, they no longer hesitated. By the time parents were interviewed, few expressed current stigma towards morphine and several said they would advise other parents to administer it more. It is unlikely parents would have been willing to admit to and identify historical negative attitudes toward pharmacological interventions, had their current attitudes also been negative. Secondly, qualitative literature is convergent regarding parents' positive attitudes towards pharmacological interventions. Although dated, one study found parents of children with cancer in Sweden denied refusing opioids to children for fear of addiction (Ljungman *et al.*, 2006). More recently, Taiwanese parents of children with neuroblastoma were found to administer more pharmacological interventions as they progressed through treatment (Lu *et al.*, 2011). Thirdly, parents provided detailed descriptions of alternative causes of their hesitation to administer pharmacological interventions: the analgesic context, and children refusing pharmacological interventions. These practical barriers present the most likely explanation for between-method divergence and will be discussed next.

### **8.5.2 *Practical barriers to pharmacological interventions***

A potential explanation of between-method divergence is that surveys may be measuring practical rather than attitudinal barriers to pharmacological interventions. Two practical barriers to pharmacological interventions hindered parents' management of their children's cancer pain at home. Firstly, the analgesic context of cancer-pain management (Section 1.4.1) means administration of analgesic drugs is often not considered a safe option for parents. The antipyretic effect of paracetamol means its administration may mask a raised temperature which, at home, could be parents' only indication of a potentially fatal infection (Oberoi *et al.*, 2013). In interviews, parents expressed fear of administering paracetamol because of this risk. Some parents reported that they had been told by HCPs, they were not allowed to administer paracetamol, even if their



child's temperature was within safe parameters. This fear has been reported in interviews with parents of children with acute lymphoblastic leukaemia on treatment or 12 months post-treatment (Bettle, 2015).

Due to its association with bleeding, ibuprofen is contraindicated for children with cancer (Cheng and Tattermusch, 2014; Hanmod and Gera, 2016). In interviews, one parent reported occasional ibuprofen administration and a few parents mentioned not being able to administer ibuprofen but were not aware of why. No parents recorded the administration of ibuprofen in pain diaries. Despite codeine being removed from drug formularies for children several years ago for safety reasons (MHRA, 2013), data in the pain diaries indicates that one dose of codeine was administered to one child. In the interviews, two additional parents, whose children had relapsed, reported administering codeine to their child prior to it being removed from formulary. Although parents did not report on its efficacy, evidence of codeine administration prior to its removal from formularies suggests it may have historically held analgesic value to parents managing children's cancer pain.

Morphine is associated with side-effects of constipation and nausea (Cheng and Tattermusch, 2014; Hanmod and Gera, 2016). The research presented in this thesis found many children were already on antiemetics and laxatives due to side-effects of chemotherapy causing constipation and nausea (Section 6.4.4.2.2). Interviews and pain diaries reveal constipation to be a frequent source of pain for many children. Parents were subsequently hesitant to administer morphine due to a fear of it exacerbating existing symptoms. Over time, some parents learned to mitigate side-effects of morphine with other interventions such as antiemetics, laxatives, and dietary changes, but many expressed a frustration with morphine causing more problems than it solved. Although in one study parents and children reported nausea, vomiting, and constipation as side-effects of opioids (Ljungman *et al.*, 1999), there is limited other evidence in literature regarding how this impacts on parents' pain management.

Compared to children who experience pain for other reasons, this analgesic context for children with cancer may have altered parents' MAQ responses. For example, parents who agree with the statement "pain medication has many side-effects", are considered to be expressing a negative attitude. Similarly, parents who disagree with the statement "there is little need to worry about side-effects from pain medication" are considered to be expressing a negative attitude. Although masking a raised temperature is not medically classified as such, given its potentially fatal consequences, many parents may consider it to be a side-effect. If parents consider masking a

raised temperature to be a side-effect of paracetamol, it is unsurprising that many would show negative attitudes to these statements. This antipyretic quality of paracetamol may be why a strong negative correlation was found when comparing the MAQ avoidance scale to paracetamol, but not morphine administrations. It may be that the MAQ is detecting practical barriers, rather than attitudinal barriers. This may explain why parents in this study seem to show more negative attitudes on the MAQ than non-cancer populations where the analgesic context is different (Twycross *et al.*, 2015d; Zisk *et al.*, 2007a, 2010) (Appendix 13).

Secondly, research in this thesis found children often refused pharmacological interventions due to finding them unpalatable (Section 6.4.4.4.3). Parents reported that when they wanted to administer pharmacological interventions they had to weigh up whether the benefit of doing so was worth upsetting or arguing with their child. Children refusing pharmacological interventions as a barrier to parents' management of cancer pain has been reported elsewhere (Bettle, 2015; Fortier *et al.*, 2014; Mariyana *et al.*, 2018). In one study, mothers of children with cancer described this as a major challenge which was time-consuming (Clarke *et al.*, 2005).

In summary, the analgesic context combined with children refusing pharmacological interventions meant parents felt they had an "empty toolbox" of pain management interventions. This led to frustration, helplessness, an inability to manage their child's pain, and feelings of being stripped of parenting skills. Paediatric cancer pain guidelines which may shed light on this area are currently being written but have not yet been published (Twycross, 2018). A literature review into children's cancer pain concluded that research is conflicted with regards to parents' attitudes towards analgesic drugs (Twycross *et al.*, 2015b). Findings from the integration of datasets reported in this thesis can explain conflicts found elsewhere in literature. Due to the analgesic context for children with cancer and because these children find pharmacological interventions unpalatable, studies which have used the MAQ may be detecting the practical barriers to pharmacological interventions which continue throughout the cancer trajectory. Conversely, interviews are able to distinguish between attitudinal barriers which resolve throughout the cancer trajectory and practical barriers which are resolved through administration of non-pharmacological interventions. This research suggests the MAQ may not be differentiating between attitudinal and practical barriers to analgesic drug administration.

### **8.5.3 Importance of non-pharmacological interventions for parents managing children's cancer pain at home**

Attitudinal and practical barriers to pharmacological interventions underpin the importance of non-pharmacological interventions for parents managing their child's cancer pain at home. Limitations of pharmacological pain relieving interventions and the imperative to increase use of non-pharmacological interventions has been previously documented for children's cancer in inpatient settings (Plummer *et al.*, 2017). Research presented in this thesis is the first to extend this finding to non-clinical settings. Use of non-pharmacological strategies to manage children's pain at home was seen as far more than a helpful adjunct to pharmacological interventions. Non-pharmacological interventions allowed parents to re-stock their "empty toolbox" and regain their ability to manage their child's pain at home. This essential aspect of pain management resulted in increased confidence for parents.

Evidence supporting non-pharmacological interventions in the treatment of children's cancer pain is growing. These interventions are often referred to as complementary and alternative medicines or physical and psychological interventions. Prevalence of non-pharmacological interventions for childhood cancer varies worldwide with estimates ranging from 47-67% (Diorio *et al.*, 2016). Prevalence of non-pharmacological interventions in children with cancer is high: one study found 65% of children with cancer on active treatment recruited from an outpatient clinic used at least one type of non-pharmacological intervention for various reasons, including cancer pain (Al-Qudimat *et al.*, 2011). Several literature reviews have investigated the effectiveness of non-pharmacological interventions for adult cancer pain (Bao *et al.*, 2016; Running and Seright, 2012), child cancer symptoms (Coughtrey *et al.*, 2018), and child cancer pain (Jibb *et al.*, 2015). Each literature review held some support for non-pharmacological interventions but concluded evidence to be of low quality and moderate at best. One literature review found six studies which reported improvement in physical symptoms of children with cancer (Coughtrey *et al.*, 2018). Specific to pain, one literature review found evidence for hypnosis, imagery, support groups, acupuncture, and healing touch in adults with cancer (Running and Seright, 2012). Another review found evidence to support use of psychoeducational interventions, music interventions, acupuncture plus drugs, Chinese herbal medicine plus cancer therapy, compound kuchen injection, reflexology lycopene, TENS, qigong, cupping, cannabis, reiki, homeopathy, and creative arts in adults with cancer (Bao *et al.*, 2014).

There is evidence, in addition to research reported in this thesis, for distraction (Bagnasco *et al.*, 2012; Gorji *et al.*, 2017; Helgadóttir and Wilson, 2014), massage (Post-White *et al.*, 2009b), cuddles (Friedrichsdorf, 2010; Friedrichsdorf and Kang, 2007), and heat (American Academy of

Pediatrics and American Pain Society, 2001; Conner-Warren, 1996) as pain management techniques in children with pain caused by a variety of conditions. In children with cancer, 69% of interventions utilising psychological or physical interventions lead to a decrease in pain (Jibb *et al.*, 2015). One oncology protocol advises on use of psychological (education, explanation, distraction, relaxation, hypnosis) and physical (warmth, cold, massage, physiotherapy, TENS, acupuncture) strategies to manage pain in children with cancer (Cheng and Tattermusch, 2014). This combined evidence represents an emerging evidence base to support the use of non-pharmacological interventions in reducing children's cancer pain (Post-White, 2006).

Research has reported cancer nurses struggle to find reputable sources of non-pharmacological information and often do so without knowledge of a robust evidence base (Rojas-Cooley and Grant, 2009). There is mixed evidence regarding HCPs attitudes towards non-pharmacological interventions. A literature review reported largely positive attitudes (Balouchi *et al.*, 2018), but a study with cancer nurses revealed negative attitudes and a belief that it is risky to combine non-pharmacological interventions with cancer treatments (Stub *et al.*, 2018). Nurses may benefit from education or training surrounding use of non-pharmacological interventions to minimise their own negative attitudes and misconceptions (Cırık *et al.*, 2017).

In summary, divergence between quantitatively and qualitatively gathered data suggest the MAQ may be detecting practical rather than attitudinal barriers to pharmacological interventions. Contrary to previous literature, combined mixed methods analysis of what influences parents' choice of interventions to manage their child's pain at home has found practical barriers, not attitudinal barriers, to be the main source of hesitancy to administer pharmacological interventions. Practical barriers include pharmacological interventions being frequently perceived as contraindicated as well as unpalatable to children. These barriers result in parents feeling they have an "empty toolbox" of pharmacological interventions which they restock with non-pharmacological interventions. This new knowledge would not have been uncovered had only one data collection method been utilised for this research.

## **8.6 Discussion of interventions**

This research concluded most children with cancer experience clinically significant pain at home and parents face practical barriers when responding to that pain. A logical next step for this research is the development of an intervention to support parents in managing their child's cancer pain at home. Before proceeding with the costly and lengthy process of intervention development (Craig *et al.*, 2008), it is important to increase the likelihood of producing a successful intervention

by learning from previous interventions to support parents managing children's pain at home. The published integrative review included in Chapter 3 of this thesis provides information required prior to intervention development (Parker *et al.*, 2018). This review found that interventions to assist parents of children with cancer managing their child's cancer pain at home are more likely to be effective when they: target parents directly or target nurse-parent interactions; are tailored and multifaceted; and consider analgesic drug effectiveness, pain education, pain assessment tools, and parent attitudes. Studies designed to test interventions stemming from this research are more likely to produce significant results when they have an adequate sample size as well as measures of adherence, pain behaviour, and a control measure of sedation which ensures true analgesic effect is achieved.

At the point at which the literature search was conducted, no interventions existed to support parents managing their child's cancer pain at home. Subsequently, a paper detailing the development and formative evaluation of an app to support parents managing children's cancer pain has been published (Chung *et al.*, 2018). There are several ways in which this app reflects recommendations stemming from the integrative review in Chapter 3. Firstly, the app is parent-targeted and included parents in its design. It is also multifaceted and addresses various aspects of parents' pain management. In line with recommendations from the integrative review, a tailoring algorithm was included to ensure contents matched the child's diagnosis and treatment. The pain education recommendation is met in four modules which provide parents with information regarding pain throughout cancer treatment. Information regarding non-pharmacological interventions is provided, but it is unclear whether the analgesic context of children's cancer pain at home is explicitly addressed in this module. One of these modules addresses barriers to pain reporting which will help parents in pain assessment and meet the pain assessment recommendation. Description regarding this module is short, but it appears not to be providing any pain assessment tools and does not advocate for bundled approaches to pain assessment which research described in this thesis suggest will be effective (Section 8.3.1). Although attitudes are not explicitly addressed, provision of accurate and relevant information in the modules may go some way to changing attitudes. In addition, while nurse-parent interaction is not addressed, one of the modules provides information on support services available which may improve this interaction. Chung and colleagues' intervention (2018) met many recommendations stemming from the integrative review in Chapter 3. They report positive feedback from parents on whom it was tested but to date provide no evidence as to its efficacy in reducing children's pain at home.

The integrative review in Chapter 3 concluded that parents should be central in any intervention designed to support them in managing their children's pain at home. Parent-targeted interventions

produced the highest number of interventions successful in reducing child pain at home (Parker *et al.*, 2018). Literature supports the importance of parents in managing children's cancer pain at home (Bettle, 2015; Fortier *et al.*, 2011b, 2014). Similarly, parents in research reported in this thesis described themselves as solely responsible for their child's pain management. These combined findings suggest parent-targeted interventions are most likely to be successful when applied to children's cancer pain at home. The integrative review in Chapter 3 also supported interventions targeting nurse-parent interactions which were, combined with parent-targeted interventions, most likely to be effective at increasing analgesic drug administration (Parker *et al.*, 2018). Literature emphasises the importance of nurses in preparing parents of children with cancer for discharge (Bettle, 2015; Flury *et al.*, 2011). In research described in this thesis parents report discussing use of HCPs as a source of information, as well as HCPs being an intervention available to them.

Chapter 3 suggested the reason for the difference between interventions which reduced child pain, and those which only increased analgesic drug administration, may be that many interventions did not provide analgesic drugs of sufficient strength to significantly reduce pain (Parker *et al.*, 2018). Inadequate analgesic strength meant that for some interventions an increase in analgesic administration did not lead to reduction in pain. A key finding of research in this thesis was that practical barriers limited administration of analgesics. This included the analgesic context in which paracetamol, ibuprofen, codeine, and morphine administration all have undesirable consequences, and children frequently find pharmacological interventions to be unpalatable. Interventions need to ensure adequate analgesics are provided and support parents in overcoming practical barriers to analgesic administration.

In addition to analgesic drug effectiveness, three components of parents' pain management were found to be important in successful interventions and will now be discussed (Parker *et al.*, 2018). Firstly, interventions may benefit from including an element of pain education. Pain education has been found to significantly increase pain knowledge and decrease pain in adults with cancer (Lovell *et al.*, 2014; Marie *et al.*, 2013). An intervention to support carers of adult cancer patients managing pain at end of life included a pain education component (Latter *et al.*, 2018). In this feasibility study, recruitment was insufficient to quantifiably report positive results of the intervention, but qualitative findings suggested the intervention was effective. To date, there is no research relating to formal pain education programmes for parents of children with cancer, only one intervention in the form of an app (Chung *et al.*, 2018). In a list of education topics for nurses to address with parents of children newly diagnosed with cancer, pain has been identified as essential (Rodgers *et al.*, 2018). This list was developed using expert consensus but has not been

empirically tested. Despite evidence in support of education, one review investigating parents' management of children's postoperative pain at home found education alone was insufficient to reduce pain (Chorney *et al.*, 2014). Research in this thesis found formal pain education to be only a small part of parents learning to manage their child's pain at home. Parents used a variety of sources in the learning phase of the model derived from interviews: information gained from HCPs, online resources, and their own instinct (Section 6.4.4.3.1); personal background including health, personality, profession, and parenting style (Section 6.4.4.3.2); and informal support of family, friends, and other people with cancer experience (Section 6.4.4.3.3). Interventions may need to encompass a variety of information sources.

Secondly, the integrative review in Chapter 3 revealed pain assessment tools provided in isolation were insufficient to significantly decrease pain experienced by children at home (Franck *et al.*, 2007; Kankkunen *et al.*, 2009; Unsworth *et al.*, 2007). However, when combined with other aspects of interventions, pain assessment tools may have benefits (Chambers *et al.*, 1997; Walther-Larsen *et al.*, 2016). Pain assessment tools may have been helpful in overcoming the challenges reported by parents in research described in this thesis. Learning was a phase in interview findings but use of pain assessment tools did not feature in research findings. It is likely that including pain assessment tools as part of a multifaceted intervention to help parents of children with cancer manage their child's pain at home will enhance the interventions' efficacy. As discussed in Section 8.3.1, any interventions will need to account for the way in which research in this thesis has found that parents assess their child's cancer pain at home and may be enhanced by including several elements of pain assessment, known as bundled approaches.

Thirdly, literature widely supports findings of the integrative review in Chapter 3 which suggested parents' attitudes towards pain medications impact on parents' pain management (Forgeron *et al.*, 2006; Fortier *et al.*, 2012; Twycross *et al.*, 2015d; Zisk *et al.*, 2010). Quantitatively, research in this thesis found parents held many negative attitudes towards analgesic drugs. Negative attitudes were also apparent in the qualitative interview data, but frequently reduced when parents realised the benefits of administering pharmacological interventions. Negative attitudes did not appear to be a deciding factor in analgesic drug administration. Nevertheless, interventions which address parent attitudes may lead to the swifter resolution of negative attitudes.

Chapter 3 suggests researchers are more likely to be successful in intervention development if they ensure the intervention: is parent-targeted; includes effective analgesic drugs; provides parents with pain assessment tools and pain education; and addresses parents' attitudes (Parker *et al.*, 2018).

Such an intervention would be considered a complex intervention (Campbell *et al.*, 2000; Craig *et al.*, 2008). Following the Medical Research Council's guidance on developing and evaluating complex interventions (Craig *et al.*, 2008) will be essential for producing an effective intervention. This guidance advocates for a cyclical process including development, feasibility/piloting, evaluation and implementation (Craig *et al.*, 2008). Feasibility and pilot testing will enable calculation of sufficient sample size for evaluation to be adequately powered (Craig *et al.*, 2008; Parker *et al.*, 2018). The evaluation phase may benefit from including control measures of sedation to ensure true analgesic effect is achieved, pain over time, and adherence (Parker *et al.*, 2018). In conclusion, findings of the integrative review described in Chapter 3 are convergent with findings of research described in this thesis and evidence from wider literature. It is important that interventions developed are parent focused and include attempts to counter practical barriers to pharmacological interventions.



## **8.7 Discussion of findings in light of the Theory of Planned Behaviour**

Section 4.3 outlines the TPB which was used to guide this research. As displayed in Figure 4.1, according to TPB, attitudes, subjective norms, and PBC contribute to intention, which leads to behaviour (Ajzen, 1991). Considering the TPB provides further understanding of parents' management of children's cancer pain at home and consideration of the findings also provides further understanding of TPB. Both considerations will be delineated in this section.

### ***8.7.1 Parental assessment of children's cancer pain at home***

Figure 8.2 presents pictorial representation of how TPB (Ajzen, 1985) can be used to understand parent assessment of children's cancer pain at home. When assessing their child's pain, TPB states parents' attitudes resulted from behavioural beliefs and outcome evaluations. The research presented in this thesis found parents expressed behavioural beliefs that actively learning to know their child's individual pain expression would result in being better able to manage their child's pain. This was coupled with parents valuing highly the outcome evaluation in which their child had no pain. Subjective norms, which consisted of normative beliefs and motivations to comply, did not appear influential in parents' pain assessment behaviour. Parents expressed feelings of being alone in their responsibility to care for their child's pain which led to subsequent pressure to alleviate their child's pain on their own. This meant they may not have felt that others valued their pain assessment behaviour as much as they personally did. As a consequence, motivation to comply with others in this context appeared less relevant. Control beliefs and control frequency, which constitute "perceived behavioural control" (PBC), did impact parents' pain assessment. Parents had control beliefs that actions taken to actively learn about their child meant they were able to adequately assess their child's pain. They acknowledged control frequency and described barriers to pain assessment behaviour by describing reasons their child would try to hide their pain which constitute display rules. Parents were able to overcome these barriers by learning to know their child's unique pain expression.

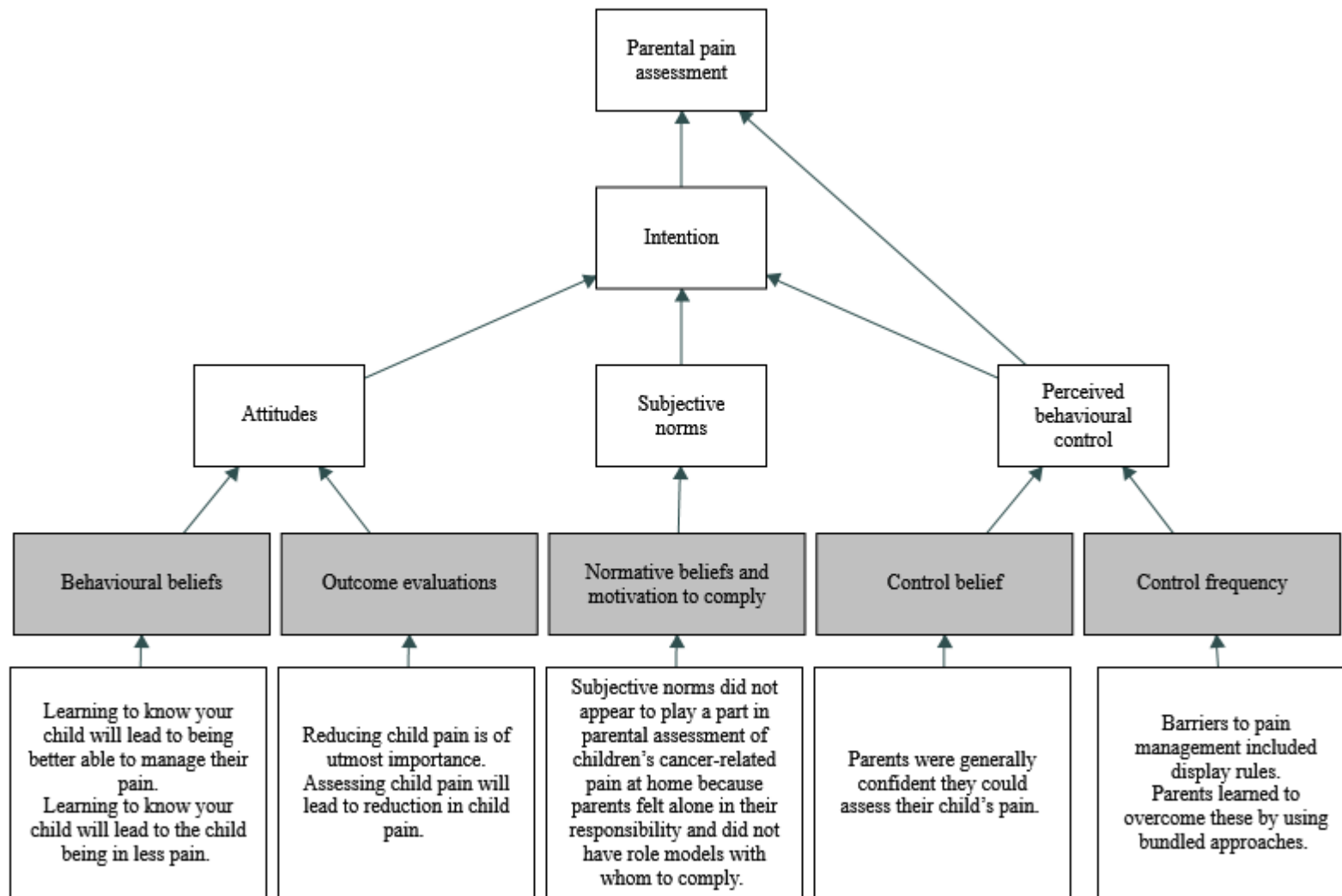


Figure 8.2: Implications of TPB on parents' assessment of children's cancer pain at home

### 8.7.2 *Parents' management of children's cancer pain at home*

Figure 8.3 illustrates how the TPB (Ajzen, 1985) can be used to explain reasons for parents' choice of interventions for managing children's cancer pain at home, as well as ways in which research described in this thesis does not align with the theory. In this research, attitudes were measured quantitatively in surveys and explored qualitatively in interviews. Integration revealed that parents had behavioural beliefs that administering pharmacological interventions may improve pain but may cause negative consequences: if parents administered paracetamol they could mask a child's raised temperature; if parents administered morphine, they could cause or exacerbate symptoms of nausea or constipation. Parents held additional behavioural beliefs that administering pharmacological interventions could result in distress for their child who found them unpalatable. Parents' outcome evaluations meant they frequently valued the outcome of non-pharmacological interventions above the outcome of pharmacological interventions.

Subjective norms, consisting of normative beliefs and motivations to comply (Ajzen, 1985), were explored qualitatively in interviews. Although questions in the interview schedule were formulated to address subjective norms, generally parents did not discuss either normative beliefs, or motivations to comply in their responses. Parents described social relationships in the form of a network of support (Section 6.4.4.3.3), but this support did not influence their pain management choices. Due to every child being different in terms of pain management needs, even other parents of children with cancer could not provide a norm with which to compare or comply. Indications of the potential for subjective norms to influence parents may be evident in qualitatively measured attitudes towards pharmacological interventions. The excerpt below (taken from Section 6.4.4.4.3) is an example of participants being aware of, but not influenced by, subjective norms.

*"...a lot people don't like using medicines. But at the end of the day, why make your child suffer? Just use medication." – Ruth*

Ruth perceives a negative public opinion of pharmacological interventions but believes this does not influence her behaviour. This type of response was typical for participants in this research.

Perceived behavioural control (Ajzen, 1985) was explored qualitatively in interviews. Parents' control beliefs in relation to pain management fluctuated throughout their child's cancer trajectory. At times, parents felt they had an "empty toolbox" which led to a distressing situation in which they could not control their child's pain. Over time, they learned how to manage their child's pain using non-pharmacological interventions which improved their control beliefs. Control frequency was limited by barriers to pharmacological interventions, such as paracetamol masking a child's raised temperature, morphine exacerbating or causing constipation and nausea, and children refusing pharmacological interventions because they found them unpalatable.

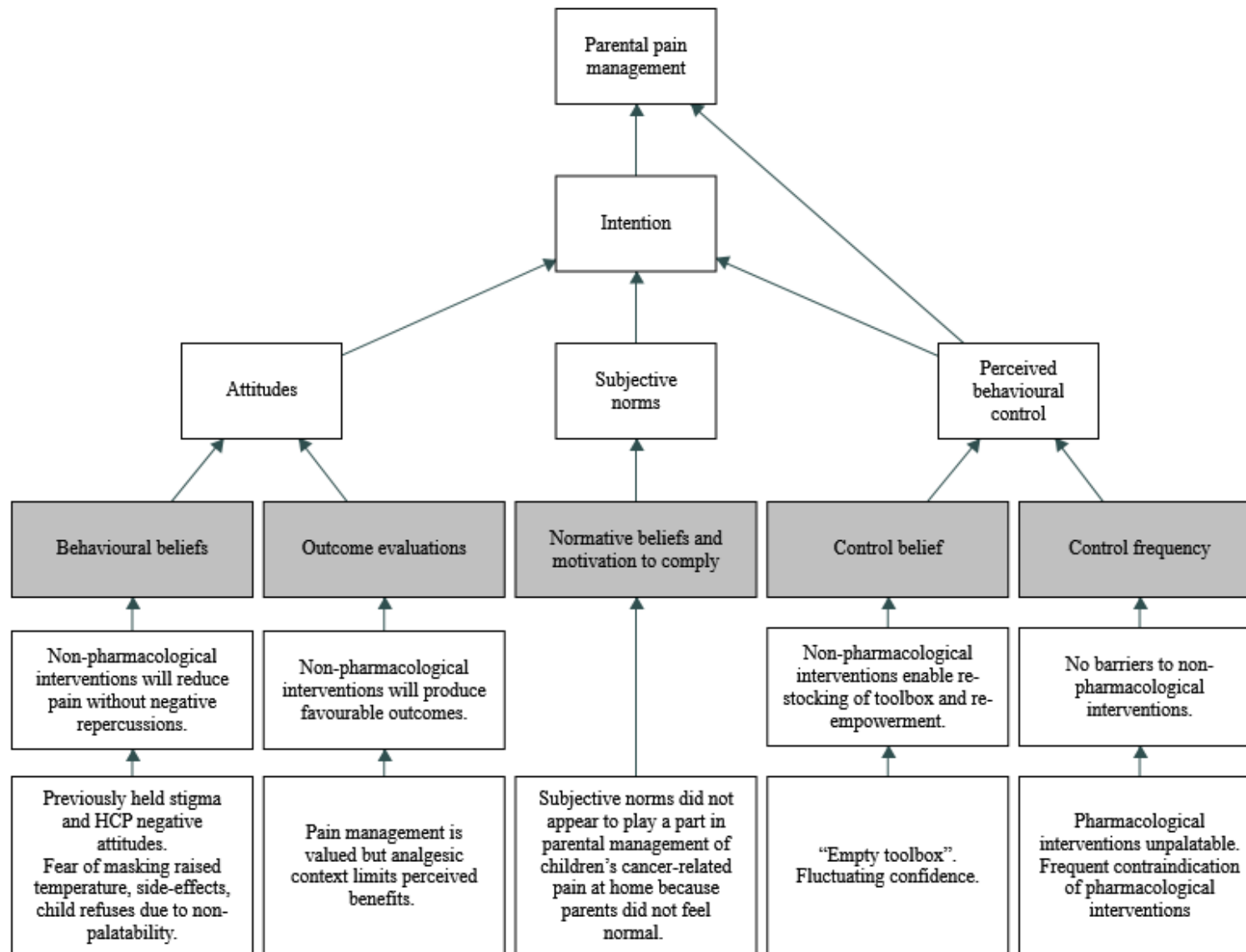


Figure 8.3: Implications of TPB on parents' response to children's cancer pain at home  
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There were fewer barriers to control frequency when administering non-pharmacological interventions. In parents of seriously ill children, perception of their child's pain being under control has been linked to confidence (Byrne *et al.*, 2011). A recent study of parents of inpatient and outpatient children with cancer both on and off treatment, found a negative relationship between parents' confidence and pharmacological interventions (Tutelman *et al.*, 2018). Tutelman and colleagues (2018) suggested this finding may be due to self-efficacy, a concept closely linked to PBC (Section 4.4) increasing as parents take a more active role in pain management. Empowering parents in pharmacological interventions increases their confidence (Cohen *et al.*, 1997; LaMontagne *et al.*, 1999; Melnyk *et al.*, 2004) and may increase PBC. In summary, behavioural beliefs, outcome evaluations, control beliefs, and control frequency each led parents of children with cancer to utilise non-pharmacological interventions with greater frequency than pharmacological interventions.

### ***8.7.3 Use of alternative theory to explain the extent to which TPB can be used to explain findings***

Although, as described in previous sections, both attitudes and PBC are clearly exposed by analysis of findings, subjective norms are not. There are three potential reasons for this. Firstly, despite formulating interview questions to address subjective norms, this method of data collection may not have been effective at deriving this information. The quote presented in Section 8.7.2 provides evidence that this is not the case: in this quote participants acknowledge the existence of subjective norms but reject potential influence of subjective norms on their behaviour.

Secondly, TPB may be unable to provide understanding of how behaviour can be explained in a context where actors do not consider themselves to be "normal". One interpretation of why subjective norms did not appear to play a significant part in either parents' pain assessment or parents' management of children's cancer pain at home may stem from feelings of being alone and helpless. Participants did not feel like "normal" parents, which may be why subjective norms did not appear relevant. Parents found themselves in a "new normal" where they did not know what others would expect them to do and had no models of behaviour from which to draw subjective norms evidenced in Section 6.4.4.3 of interview results. Studies have reported families of children with cancer striving for normalcy (Flury *et al.*, 2011; Jibb *et al.*, 2018) but reporting its loss (Molinaro and Fletcher, 2018; West *et al.*, 2015). This facet of TPB may not be applicable in situations where families feel they are not "normal", including families of children with cancer and other long-term conditions (Huby *et al.*, 2017; Rehm, 2013; Smith *et al.*, 2015).

Thirdly, other theories may provide understanding as to why subjective norms appear not to be relevant. Maslow’s hierarchy of needs provides a potential explanation for the lack of subjective norms in parents’ management of children’s cancer pain at home (Maslow, 1943; Maslow and Lewis, 1987). Maslow (1943) postulates that human motivation is a product of five hierarchically organised sets of goals: physiological, safety, love, esteem, and self-actualisation. Humans aim to reach the first goal, but do not aim to reach subsequent goals until basic needs within the first goal have been met. This theory has been applied by palliative care researchers who adapted each goal to their specific setting (Zalenski and Raspa, 2006). Table 8.2 shows how Zalenski and Raspa (2006) mapped Maslow’s hierarchy of needs to the palliative care context.

Table 8.2: Comparison of Maslow's hierarchy of needs with a palliative framework equivalent

<b>Maslow’s hierarchy of needs</b>	<b>Zalenski and Raspa’s palliative framework</b>
Physiological	Distressing symptoms such as pain or dyspnoea
Safety	Fears for physical safety, of dying or abandonment
Love	Affection, love and acceptance in the face of devastating illness
Esteem	Esteem, respect, and appreciation for the person
Self-actualisation	Self-actualisation and transcendence

The most basic need in this setting is the management of distressing symptoms such as pain. Devotion and effort were evident in the learning phase of interview results (Section 6.4.4.3), pressure was resultant from the context phase (Section 6.4.4.2.4), and outcomes of each pain episode affected parents’ confidence (Section 6.4.4.5.3). This indicates pain management was a high priority for parents. Social needs, which relate to subjective norms, including love, acceptance, esteem, respect, and appreciation do not feature until pain management has been achieved. This comparison, combined with research presented in this thesis, has relevance for TPB as it may explain why subjective norms, which are social in nature, did not influence either pain assessment or pain management. It may be that parents are focused on the most basic of needs, such as removing their child’s pain, and therefore do not attend to higher level social needs. As a result, normative beliefs and motivation to comply do not feature in deciding parents’ pain management behaviour.

The Theory of Planned Behaviour states that PBC can directly influence behaviour without leading to intention first (Ajzen, 1985). This aspect of TPB was not seen in analysis or research findings presented in this thesis. The research described in this thesis investigated PBC using interviews and behaviour was measured using pain diaries. There was no single data collection method which

attempted to measure the influence of PBC on behaviour and so the extent to which it exerted a direct effect on behaviour unmediated by intention, cannot be defined.

#### 8.7.4 Interventions to support parents managing children’s cancer pain at home

The TPB (Ajzen, 1985) has been valuable in understanding parents’ management of children’s cancer pain at home and interventions may benefit from use of this theory in design. Interventions supported by theory are more likely to be successful (Aldiss *et al.*, 2015). This section will consider how TPB can be used to understand findings of the integrative review in Chapter 3 which investigated interventions to support parents managing children’s pain at home. Each key integrative review recommendation will be considered in relation to TPB including: parent-targeted, nurse-parent directed interventions, attitudes, pain education, pain assessment tools, tailored interventions, multifaceted interventions, and effective analgesic drugs. Table 8.3 shows how findings from the integrative review map to the TPB.

Table 8.3: Findings of literature review mapped to TPB

Theory of Planned Behaviour	Findings from integrative review	
Attitudes	Attitudes	Parent-targeted Multifaceted Tailored
Subjective norms	Target nurse-parent interactions (but diverges from other thesis findings)	
Perceived behavioural control	Pain education Pain assessment	

Firstly, TPB and the integrative review converge on attitudes being part of improving parents’ management of children’s pain at home. Secondly, the integrative review suggested targeting nurse-parent interaction, relating to the social aspect of TPB known as subjective norms, may improve analgesic drug administration. Findings described in other areas of this thesis suggest subjective norms may not be relevant for parents of children with cancer (Sections 8.7.1, and 8.7.2). None of the studies included in the integrative review related to parents of children with cancer, so this aspect of congruence with TPB may not be relevant for interventions designed for this population. Thirdly, the integrative review and TPB converge on the importance of PBC in improving parental pain management at home. The integrative review advocates for pain education and inclusion of pain assessment tools as facets of interventions which are likely to improve parents’ management of children’s pain at home. Both pain education and pain assessment tools are likely to improve control beliefs by empowering parents to believe they are able to manage their

child's pain at home. Equally, pain education and pain assessment are likely to improve control frequency and reduce parents' perception of barriers to pain management.

Three findings from the integrative review map to the TPB as a whole. Firstly, interventions are more likely to be successful if they are parent-targeted. This relates to every aspect of TPB which is a behavioural theory focusing on the individual. The integrative review adds support to TPB by finding that interventions which address the individual whose behaviour it aims to change are more likely to be successful. Secondly, interventions are more likely to be successful if they are multifaceted. Here the TPB can support the integrative review by providing guidance about what each facet of the intervention should include. In the same way that TPB postulates changes in one concept alone is insufficient to produce change, so the integrative review postulates that interventions which address one aspect of parental pain management are unlikely to be successful. Thirdly, interventions are more likely to be successful if they are tailored. Although this relates to every aspect of TPB, there is nothing within the key concepts of TPB to suggest the importance of individualisation of interventions. Perhaps this divergence with TPB can contribute to understanding of TPB in future research. One finding from the integrative review, the importance of analgesics of adequate strength, has not been discussed in relation to TPB as it does not relate to behaviour, but rather to pharmacology. In summary, many aspects of TPB can be linked to findings from the integrative review. Where findings diverge, this provides new knowledge in understanding of TPB. This analysis has revealed insight into what type of interventions are likely to be successful in supporting parents to manage their child's cancer pain at home.

## **8.8 Strengths and limitations**

This section will discuss aspects of the methods and sample in this research which relate to validity, credibility and trustworthiness of results. Initially each data collection method will be discussed. Following this strengths and limitations of integration in this research will be highlighted.

### **8.8.1 Survey processes**

#### **8.8.1.1 Participant attrition**

Examination of the screening log compared to demographics of participants who completed surveys revealed that the sample population was not statistically significantly different from the potential participant population in terms of child gender, child age or parent ethnicity. No examination of hospital-held patient records was used in completion of the screening log which limited demographic information available. It cannot be determined whether other demographic



variables such as ethnicity, socioeconomic status, or education may have influenced choice to participate. Despite this, there is no indication that data collection methods hindered participation of individuals based on variables of data collected.

#### **8.8.1.2 Sample characteristics**

In this study, nearly twice as many parents of boys were recruited than parents of girls (60 and 36 respectively). Childhood cancer incidence in the UK population is slightly higher for boys than girls (1/410 and 1/471 respectively) (Irvine, 2017). In addition, boys with leukaemia have longer treatment than girls which may have resulted in increased recruitment opportunities (Children with Cancer UK, 2018). Although this somewhat accounts for the gender imbalance, it is unclear why it was so marked. As there was no statistically significant difference in the proportion of boys whose parents decided to participate and those who chose not to participate, there is no indication that this disproportion should be attributed to recruitment strategy or method of data collection.

Age of children in the sample population roughly reflects the UK population where childhood cancer incidence is highest for children aged from birth to five years old, reduces from five to nine years, and slightly increases from age ten (Irvine, 2017). Data collection for this study was conducted in a hospital which does not provide for children under one year of age. Participants were recruited from an outpatient unit for patients up until 24 years of age and a children's inpatient ward for children from 1-15 years of age. Data collection did not take place on the teenage inpatient ward which provides for patients age 15-24 years. Consequently, although children were eligible for this research up until the day before their 17th birthday, the location of data collection explains the absence of parents of children under one year of age and may have resulted in fewer parents of 15 and 16-year-old children sampled. Leukaemia is the most frequent childhood cancer diagnosis in the UK making up 30% of diagnoses (Irvine, 2017). High prevalence of leukaemia in this sample (55%) may be due to frequent outpatient appointments and duration of treatment required by leukaemia treatment protocols creating multiple sampling opportunities. Distribution of remaining diagnoses roughly matches the UK population (Irvine, 2017).

The negative effects of excluding non-English speaking participants in research has long been documented (Frayne *et al.*, 1996) and the risk that this practice biases the research base as a whole is a continued concern (Rasmussen and Montgomery, 2018). Ethnic background of the survey sample is comparable to the 2011 census where 86% of the UK population was white (65% child, 70% parent in this sample), 7.5% Asian (11% child and parent in this sample), 3.4% Black (8% child, 12% parent in this sample) and 2.2% Mixed (12% child, 3% parent in this sample) (Office

for National Statistics, 2012). Seventy-three percent of participants were mothers, reflective of participation in similar studies (Bettle, 2015; Fortier *et al.*, 2012, 2014; Zhukovsky *et al.*, 2015) and the demographic of carers more widely (Family Caregiver Alliance National Center on Caregiving, 2015). Forty-five percent of children had been diagnosed less than six months prior to recruitment. During these first six months, hospital visits are frequent creating multiple sampling opportunities.

Convenience sampling was used for recruitment to this study for both the survey and pain diary. This strategy allowed the possibility that the sample of participants who expressed interest in completing an interview may be biased and the sample should not be considered generalisable (Parahoo, 2014). In addition, this strategy led to a varied sample with a wide range of time since diagnosis, different diagnoses, and ages. However, this sampling strategy facilitated recruitment of a sample size sufficient for statistical analysis (Etikan *et al.*, 2016; Parahoo, 2014). Participants were recruited from just one UK tertiary cancer centre which limits generalisability. Due to the organisation of UK paediatric cancer services, children with cancer will be allocated one tertiary cancer centre which conducts specialist aspects of treatment and one shared care centre which conducts general aspects of treatment closer to home (NHS England, 2017). This recruitment strategy was advantageous as it meant that this research includes representation from parents whose children are receiving care from a range of shared care centres.

Recruitment relied on HCPs acting as gatekeepers to ensure potential participants were not approached at times when recruitment may have incurred additional distress (Creswell and Plano Clark, 2011; Teddlie and Tashakkori, 2009). This strategy was a safety mechanism designed to reduce distress and overcome a potential ethical barrier. However, it is not possible to say definitively whether HCPs fully understood potential participants' needs in these circumstances or the impact of research on participants. Indeed, paternalistic HCPs may have prevented potential participants from experiencing potential benefits of involvement in research (Coombs *et al.*, 2016).

## **8.8.2 Pain diary processes**

### **8.8.2.1 Participant attrition**

Examination of demographic criteria for participants who completed surveys compared to participants who completed pain diaries revealed that the sample population did not differ from the potential participant population in terms of child gender, child age, or parent ethnicity. Due to risks of obtaining significant results by chance alone when conducting many statistical tests, no other demographic variables were studied in relation to attrition, so it has not been determined whether other demographic variables may have influenced choice to participate. However, there is no

indication that data collection methods hindered participation of individuals based on variables which were investigated.

### **8.8.2.2 Sample characteristics**

Child demographic characteristics of gender, age, diagnosis, and ethnic background for pain diary sample were similar to that of the survey sample. Parent ethnicity and relationship to child were also distributed similarly to the survey sample.

### **8.8.2.3 Data quality**

Most parents preferred to complete paper copies of the pain diary. Compliance was higher with paper copies and parents reported anecdotally that having something physical served as a reminder. Data quality for pain diaries was low with some fields having inadequate data to analyse statistically. Parents rarely completed outcome data, and frequently omitted data from other fields. Compliance for pain scores were relatively high with a mean of 82% of episodes including a pain score. Previous research has found non-compliance to be an issue in real-time diary style data collection (Kearney *et al.*, 2009). Using incentives and researcher reminders, 97.9% completion has been achieved in a pilot study of pain diaries in children with cancer (Fortier *et al.*, 2016). Research described in this thesis encouraged completion of pain diaries by meeting with participants prior to pain diary commencement with the purpose of ensuring participants understood their contribution. Although low quality data prevented some statistical analyses, the amount of data provided were sufficient for useful inferences to be drawn in terms of pain score, location, cause, and parent response to pain. In addition, data were adequate for inferential statistics to be performed on aggregated datasets compared to survey results.

Previous research provided parents with a list of responses for each facet of pain (Madi and Clinton, 2018; Tutelman *et al.*, 2018). In the research described in this thesis, parents were not provided with a list to choose from but were free to respond with whatever they perceived to be the cause of their child's pain. This meant parents' responses were not influenced or limited by options available which has two implications for the outcome of this research. Firstly, this may have led to reporting inconsistencies. For example, if chemotherapy toxicity caused mucositis for two children, one parent may have recorded the cause of pain as chemotherapy toxicity, and another as mucositis. This potential for reporting inconsistencies shows the value of establishing a standardised list of causes of pain for children with cancer at home. Secondly, as parent responses were not standardised, an element of researcher interpretation was required in categorising of responses. Thirdly, this research can provide new knowledge to this body of literature by providing

a list of causes of pain in children with cancer at home. Although extensive, this list does not claim to be complete and further research will be required before it can be standardised. Importantly, this list was developed directly from parents, with limited influence from HCPs (Appendix 14).

#### **8.8.2.4 Use of proxy reports of pain**

Not including a self-report measure of pain means pain measures in this research can only be considered an estimate of children's pain (Zhou *et al.*, 2008). Using parents as participants matched the research aim of understanding how parents of children with cancer manage their child's pain at home. This research intentionally focused on parents as they increasingly take responsibility for pain management from HCPs (Fortier *et al.*, 2011b). The integrative review (Chapter 3) identified parent-targeted interventions as most likely to reduce child pain at home adding further evidence that a focus on parents was appropriate (Parker *et al.*, 2018). However, this focus on parents may have a detrimental effect on a child's right to be involved in their own care (Coyne *et al.*, 2014; Rost *et al.*, 2017; Ruhe *et al.*, 2016). There is growing recognition of the importance of the child's voice in clinical and research settings (Söderbäck *et al.*, 2011). Children with cancer have expressed a desire to become more involved in their own care, especially as they get older (Darcy *et al.*, 2014; Gibson *et al.*, 2010) and report a sense of being overlooked (Ruhe *et al.*, 2016). Involvement of children in future research will enhance child participation in their own pain management at home with parents.

It was appropriate to use parents to rate their child's pain for several reasons. Firstly, Section 8.3.1 describes potential inaccuracies which may have arisen had a self-report measure of pain been used in this research. Children's use of display rules, such as not admitting to pain due to not wanting to be given a pharmacological intervention (Section 8.3.1), may have prevented them from accurately reporting their pain (Versloot *et al.*, 2013). Secondly, parents describe using many, if not all, aspects of CARES bundled approaches recommended by expert researchers (Twycross *et al.*, 2015c), leading research in this thesis to conclude that parents are able to accurately assess their child's pain at home. Thirdly, using parents to rate their child's pain ensured every child was represented regardless of their communication abilities (Hedén *et al.*, 2013). In summary, although the absence of a self-report measure means certainty of assessment is not guaranteed, using parents to report their child's pain was appropriate and likely to be of sufficient accuracy for the conclusions drawn.

### **8.8.3 Interview processes**

#### **8.8.3.1 Sample characteristics**

Purposive sampling was used for the interviews, which allowed data to be gathered for a broad range of experiences related to the research questions (Heavey, 2014). Participants were selected to meet the sampling frame which ensured an even spread of ages and time since diagnosis. With a single exception, criteria to meet the sampling frame were met. As with the survey and pain diary samples, a majority were mothers and most frequent diagnosis was leukaemia.

#### **8.8.3.2 Generalisability and rigour**

Polit & Beck (2010) discuss three types of generalisability: statistical generalisability which quantitative research traditionally strives to achieve; analytical generalisability in which the researcher, at point of analysis and interpretation, identifies inferences with relevance to whole sample; and transferability, or case-to-case translation. This research did not aim to achieve statistical generalisability, but instead put strategies in place to enhance analytic generalisability and transferability. Transparency over the limitations of inferences made helps readers identify the trustworthiness of findings. In addition, use of quantifiers for example, “many”, “some”, and “few”, in reporting qualitative data helps readers understand the strength of opinions expressed in this research and increased analytic generalisability (Sandelowski *et al.*, 2009). Rich description of the sample has been provided to facilitate transferability (Polit and Beck, 2010).

Interviews were conducted following the procedures outlined in Section 5.7.3. To increase rigour, questions were open ended, vague answers were clarified, and each interview ended with a review of the interview and opportunity for participants to clarify any misunderstanding (Lu *et al.*, 2011). Participants frequently used this opportunity which suggests it is a helpful mechanism for increasing rigour. Reflexivity was increased by keeping a reflective journal throughout as well as holding regular supervisions (Teddlie and Tashakkori, 2009).

#### **8.8.3.3 Assessment of subjective norms**

The interview schedule was designed to assess PBC and subjective norms. Perceived behavioural control was evident in interview findings but subjective norms did not appear to feature. It may be that interview questions and analysis process were either insufficiently nuanced or not specific enough to detect subjective norms as an influence on participant behaviour. Individuals may often be unaware of the extent to which they are influenced by others, making this concept challenging to investigate (Ajzen, 1985). However, several considerations suggest this may not be the case.

Firstly, research in this thesis suggested supplementing TPB with Maslow’s hierarchy of needs may

offer further explanation of the lack of subjective norms in findings (Section 8.7.3). Secondly, parents described the absence of normality and the need to learn a “new normal” (Section 6.4.4.3) which provides a potential explanation for the absence of subjective norms. Finally, analysis of interview results with specific attention to subjective norms revealed that participants were aware of this potential influence but did not allow it to dictate their behaviour (Section 8.7.2). It cannot be concluded for certain that subjective norms did not contribute to parents’ management of children’s cancer pain at home but these considerations suggest that subjective norms were not an influence.

#### **8.8.4 Mixed methods integration**

Each data collection method was designed to meet a different facet of the same phenomena: parents’ management of children’s cancer pain at home. For each participant, data collection began with surveys, then pain diaries, then interviews. Ordering of data collection methods was the same for every participant, so influence of one data collection method over subsequent data collection methods cannot be ascertained. It is unknown how surveys may have affected pain diaries and interviews. However, this pragmatic decision meant firstly, surveys could be used to give participants a flavour of participation in research before agreeing to more time costly data collection methods which aided recruitment. Secondly, completion of pain diaries prior to interviews meant pain diary data could be used to facilitate interviews by aiding parents’ memory and providing substance for parents to reflect on. Had pain diaries not been part of interview data collection, parents would have been reliant purely on their own recall which has been shown to be less accurate than real-time data collection (Noel *et al.*, 2010). Parents frequently commented on how useful pain diaries were in assisting their recall of pain episodes. Thirdly, this ordering removed the risk that the Hawthorne effect may mean interviews had an impact on how participants completed surveys and pain diaries (Todd, 2010).

Results of mixed methods research are more than the sum of their parts (Guetterman *et al.*, 2015). Table 5.1 shows how each data collection method contributed to the aim and research questions. This led to an enriched understanding of the phenomena which would not have been possible had any one of the data collection methods not been included (Bryman, 2006; Dickson *et al.*, 2011). This research used a mixed methods design with one primary purpose – complementarity (Greene *et al.*, 1989) – and three secondary purposes – offset (Petros, 2011), completeness (Bryman, 2006), and explanation (Bryman, 2006) – each of which strengthened the study. In many areas, datasets converged which facilitated meta-inference generation. However, it should not be assumed that convergence means datasets offer the correct finding as it is possible for more than one data collection method to have the same limitation (Moran-Ellis *et al.*, 2006; O’Cathain and Thomas, 2006).

Complementarity strengthened the study as illustrated in Table 5.1: each method examined a different facet of parents' management of children's cancer pain at home and findings complemented each other. Using a mixed methods sampling strategy (Teddlie and Yu, 2007) strengthened this study by ensuring strengths and weaknesses of different methods offset one another (Bryman, 2006; Petros, 2011) (Section 5.2.3). Depth of data was obtained from a small qualitative sample and breadth of data was obtained from a large quantitative sample (Creswell and Plano Clark, 2011). This sampling strategy provided flexibility which allowed parents to match participation to their time capacity and interest for research which may have increased sample size and inclusivity (Newington and Metcalfe, 2014). Survey and interview sample targets were both met, and pain diary recruitment was only three participants shy of target. A response rate of 63% for the survey demonstrated acceptable, non-burdensome research methods which suggested the study design was inclusive. With a single exception, criteria for the purposive sampling frame used to recruit participants to interviews was met. As a result, interview data included views from a range of participants in terms of age and time since diagnosis. By using homogenous sampling (Dickson *et al.*, 2011), meta-inferences derived from integrating different methods could be made with greater authority. Experience of child pain was not an inclusion criterion for this sample which meant the sample, unlike previous samples with research in this area (Bossert *et al.*, 1996), this research was not biased towards children who had recent experience of pain. This may have led to findings of the absence of pain.

As Figure 8.1 shows, completeness was a further strength of research described in this thesis. This research does not claim completeness in the fullest sense (Bryman, 2006), but a more comprehensive understanding of parents' management of children's cancer pain at home has been obtained using mixed methods. An examination of how and why parents of children with cancer choose to intervene in their child's pain could not have been conducted without understanding what children's pain manifestation is, and how parents assess that pain. Similarly, in this research, understanding of children's pain manifestation could not be conducted without understanding how parents assess that pain.

Although not a planned purpose of the design, as analysis progressed, it became apparent that explanation was a further purpose, and therefore a further strength of the research. Explanation in mixed methods occurs when findings from one method were used to explain findings from another (Bryman, 2006). In this research, parents described practical barriers to analgesic drug administration in qualitative interviews which in part explained negative attitudes towards analgesic drugs as expressed in quantitative surveys and under-medication of children's pain as

discovered in pain diaries. Using TPB (Ajzen, 2011), it could be shown that practical barriers, found in interviews, contributed to behavioural beliefs, which led to negative attitudes towards analgesic drugs, found in surveys. In addition, consideration of outcome evaluations could be used to understand why parents preferred administration of non-pharmacological interventions in pain diaries and interviews.

A further strength of this mixed methods design was the way in which analysis was conducted to embrace, rather than ignore divergences (Creswell and Plano Clark, 2011; Teddlie and Tashakkori, 2009). Examination of divergences led to new findings in response to several research questions. Findings which stemmed from between-method divergences would not have been revealed had this research not embraced divergences arising from mixed methods. Perhaps the most important benefit of using mixed methods is that, had any one method been absent from this study, different conclusions would have been drawn. Analysing divergences between qualitatively and quantitatively measured attitudes towards pharmacological interventions revealed practical barriers which TPB (Ajzen, 2011) considered barriers to control frequency. Similarly analysing divergences in the perceived outcome of administering pharmacological interventions aided understanding of concepts of behavioural beliefs and outcome evaluations (Ajzen, 2011). This research demonstrates how mixed methods produces findings which are more than the sum of their parts (Teddlie and Tashakkori, 2009).

## **8.9 Summary**

This chapter has finalised the integration of findings using a contiguous approach through narrative. Findings are discussed in light of literature and TPB. Initially the relationship between research questions was delineated, followed by an overview of findings. This was followed by four sections which addressed each of the research questions. Each data collection method was integrated to answer the research questions and confirmatory and contradictory literature discussed. Following these four sections, the integrative review in Chapter 3 was discussed in light of thesis findings and wider literature. Using TPB, findings were illuminated and implications for understanding theoretical frameworks were described. Final sections described strengths and weaknesses in terms of each data collection process. The next chapter provides a conclusion to the thesis.



## **Chapter 9. Conclusion**

### **9.1 Overview of conclusion chapter**

This chapter begins with a summary of the findings of research described in this thesis. Contributions to new knowledge are then set out in seven sections: four sections relating to each of the research questions, one section relating to the integrative review in Chapter 3, one section relating to methodological contributions, and one section relating to TPB. Next, implications for clinical practice, future research, and policy are described. This is followed by a description of the dissemination strategy and reflexive perspectives stemming from the reflective journal kept as part of the research process. The chapter closes with concluding remarks.

### **9.2 Summary of the thesis**

This research used a convergent, parallel design to understand how parents of children with cancer manage their child's pain at home by answering four research questions. This research was strengthened by using a mixed methods design for the purposes of complementarity, offset, completeness, and explanation. Further strength was gained by embracing rather than discarding divergences in findings. This means results are more than the sum of their parts.

Initially this research considered the pain manifestation of children with cancer at home. In line with wider literature, parents perceived their child's pain to be primarily caused by treatment side-effects. The most frequent pain locations were abdomen, legs, mouth/throat, head, and bottom. Pain location can help ascertain cause and therefore treatment of pain. Heterogeneous pain trajectories presented a potential explanation for within-method divergence regarding pain prevalence. Most of the time children are not in pain at home. However, most children experience episodes of clinically significant pain. Due to the unpredictability of pain at home, it is important all parents of children with cancer are prepared for this role.

Secondly, this research asked how parents assess their child's pain at home. Divergent findings were discussed. Two parent-based explanations (that parents were inaccurate in their perception of their pain assessment abilities, and that interview participants were a biased sample better at pain assessment) were considered. Two method-based explanations (that PPEP questions may have misled parents, and that PPEP may lack sufficient nuance to detect pain assessment abilities in the context of every child being different) were considered. Analysis of these explanations concluded

that method-based explanations were most likely to be the cause of divergences. Overall, evidence suggests parents use bundled approaches to effectively assess their child's pain at home. Researchers and HCPs may benefit from using these techniques in research and clinical settings.

Parents of children with cancer frequently under-medicate their child's pain at home but instead use a variety of non-pharmacological interventions. No previous research has quantitatively measured the frequency of parents' responses including non-pharmacological interventions to manage children's cancer pain at home. Between-method divergence regarding what influences parents' choice of interventions to manage their child's pain at home is resolved by the suggestion that the MAQ may be detecting practical barriers rather than attitudinal barriers. Consideration of practical barriers to pharmacological interventions enabled further understanding of parents' choice of intervention. Paracetamol administration risks masking a raised temperature, ibuprofen is associated with a risk of bleeding, codeine is no longer recommended for children, and morphine administration risks causing or exacerbating nausea and constipation. In addition, children refused pharmacological interventions finding them unpalatable. Parents felt they had an "empty toolbox" which they re-stocked with non-pharmacological interventions. This highlights the importance of non-pharmacological interventions for parents managing children's cancer pain at home.

The integrative review described in Chapter 3 of this thesis, combined with the empirical research, bridged the gap in literature by providing practical guidance on intervention development to support parents of children with cancer when managing their child's cancer pain at home. Conclusions from this literature review mean that there is now sufficient evidence to design interventions which aim to support these parents and ultimately reduce unnecessary pain experienced by children with cancer.

Parents' pain assessment and pain management behaviours were further explained using the TPB. In relation to pain assessment, parents held behavioural beliefs that learning to know their child would increase their ability to manage their child's pain and they valued the outcome of having a pain-free child highly. Subjective norms did not appear relevant, as parents had no role models in pain assessment. Despite describing control frequency barriers, parents control belief was that they felt confident they were able to assess their child's pain. In relation to pain management, parents' behavioural beliefs were that whilst administering pharmacological interventions could improve pain, it would likely have negative consequences. Their outcome evaluation was that using non-pharmacological interventions would have a more favourable outcome. Once again, subjective norms did not appear relevant for parents managing their child's cancer pain at home. Control

beliefs fluctuated with each pain episode. Barriers to control frequency were the practical barriers of analgesic context and non-palatability of medication. Each facet of TPB led parents to choose to manage their child's pain at home with non-pharmacological rather than pharmacological interventions. This research enhanced understanding of TPB by considering why subjective norms did not feature in parental pain management. It was suggested that subjective norms may not be relevant in situations such as parenting children with chronic conditions where individuals do not feel "normal". In addition, Maslow's hierarchy of needs may be combined with TPB to explain the absence of subjective norms in parents' management of children's cancer pain at home. Specifically, parents are trying to meet basic goals for their child including pain management and therefore have not progressed to trying to attain higher-level social goals which would constitute subjective norms.

### **9.3 Contribution to new knowledge**

#### ***9.3.1 Pain manifestation in children with cancer at home***

This research constitutes the first UK study to measure pain manifestation in children with cancer at home from parents' perspectives, and the first study worldwide to measure pain manifestation in children under the age of four years. Findings confirmed a trend that pain in children with cancer is caused primarily by treatment and this research is the first to find treatment to be the primary cause of pain at home. The most frequent pain locations are similar to previous research. It is important to generate similar findings in different settings to increase generalisability of the body of literature as a whole (Firestone, 1993; Polit and Beck, 2010). Therefore, these confirmatory findings are an important contribution to literature. Previous research concluded that overall pain intensity in children with cancer at home is mild (Fortier *et al.*, 2014). Whilst this research found that most of the time, children have very little or mild cancer pain at home, it additionally contributes the finding that most children have episodes of clinically significant pain during their cancer journey. This finding was obtained by increasing the length of real-time data collection. In addition, interviews allowed data to be collected regarding pain episodes which may not have occurred during the pain diary period. This research adds new knowledge on a national and international level to the understanding of children's pain manifestation at home in terms of cause, location, and prevalence.

#### ***9.3.2 Parental assessment of children's cancer pain at home***

This research is the first to combine pain assessment in parents of children with cancer measured quantitatively and explored qualitatively. Previous quantitative research measuring assessment in parents of children with cancer found only negative results in terms of parents' misconceptions

regarding children's pain expression (Fortier *et al.*, 2012). Had this research used solely quantitative methods to measure parents' pain assessment abilities, similar conclusions may have been reached. Due to the combination of data collection methods, this research was able to add new knowledge that parents do learn to assess their child's cancer pain at home. This research was able to explain why previous studies had concluded only negative findings regarding parents' pain assessment abilities. In acute settings, parents may not have sufficient exposure to their child's pain to allow them to learn their individual pain expression. Children with cancer experience repeated pain episodes which can be described as chronic (Fortier *et al.*, 2014). In this context, quantitative measures lack sufficient nuance to accurately detect differences between children's pain expressions.

In addition, this research is the first to describe how parents assess their child's cancer pain at home. Evidence for display rules hindering disclosure of pain in children with cancer suggests researchers and clinicians utilising child self-report should consider whether display rules may limit the accuracy of self-report measures. Parents went through the learning phase to become experts in their child. They then used several different information sources, which constitutes a bundled approach to pain assessment. Parents combined context, self-report, behaviours and emotional assessment with their personal preferences, as well as knowledge of risks of pharmacological and non-pharmacological interventions. This research is the first study to report parents' use of several aspects of bundled approaches in assessment of children's cancer pain. No previous research has described parent assessment of children's cancer pain to this level of detail.

### ***9.3.3 Parents' choice of interventions to manage children's cancer pain at home***

This research makes a unique contribution to literature by describing interventions parents use to manage children's cancer pain at home, not limited to analgesic drug interventions. This research is the first to measure parents' real-time interventions in their child's cancer pain at home for a period of more than two weeks. No previous research has measured parents' non-pharmacological interventions to manage their child's pain at home in real-time. In concordance with previous studies, this research found parents were frequently not administering pharmacological interventions to their child despite clinically significant pain. Had this research investigated only pharmacological interventions, it may have concluded in line with previous research, that parents are not attending to their child's pain and children with cancer are experiencing pain at home which is being untreated by their parents. This research did not restrict parents during data collection but allowed them to describe any pain management practice and can therefore make a new contribution to knowledge: parents do respond to children's cancer pain at home and intervene using a variety of non-pharmacological interventions.

### ***9.3.4 Influencers of parents' choice of interventions to manage children's cancer pain at home***

This research is the first to examine quantitatively and qualitatively gathered data on attitudes towards pharmacological interventions in parents of children with cancer. Using quantitative methods, previous research found high levels of negative attitudes and misconceptions towards pain medication in parents of children with cancer (Fortier *et al.*, 2012, 2014). Similar conclusions may have been reached had this research used only quantitative methods. Use of both qualitative and quantitative methods meant this research was able to add new knowledge which suggests that practical barriers contribute more to parents' hesitancy to administer pharmacological interventions than attitudinal barriers. During interviews, participants expressed relatively positive attitudes towards pharmacological interventions, so it may be that quantitative methods are detecting negative opinions caused by practical rather than attitudinal barriers. Although a previous research study describes this in an inpatient setting (Plummer *et al.*, 2017), this research is the first to document practical barriers to analgesic drug administration encountered by parents of children with cancer at home. This thesis presents the first research to link the implications of morphine side-effects to parents' pain management and ultimately, pain experience of children with cancer at home. Previous research has not outlined explicitly the reasons parents may not administer paracetamol, ibuprofen, codeine, or morphine to their child at home and the extent to which this leaves parents with an "empty toolbox". Previous research has not emphasised the extent to which children with cancer find pharmacological interventions unpalatable and frequently refuse them. This research is the first to highlight the vital role that non-pharmacological interventions play in supporting parents to manage their child's cancer pain, where pharmacological interventions are of limited acceptability to both parents and children.

### ***9.3.5 Effective interventions to support parents' pain management at home***

This thesis includes the first integrative review of literature investigating interventions to support parents managing children's pain caused by any clinical condition. It is the first review to expand inclusion criteria for studies to consider all painful conditions in children rather than purely postoperative pain. This integrative review is unique in its analysis of studies from the perspective of nurse, parent, and child interaction. The results of the review provide a checklist of ways in which researchers can design successful interventions which included recommendations for the focus of interventions, characteristics of interventions, and design of studies which test interventions. The review bridges the gap in literature between the empirical investigation of research in this thesis and intervention development. Without the findings of this integrative review, research in this thesis could not lead to effective intervention development.

### ***9.3.6 Methodological contributions to new knowledge***

This research is the first to consider pain assessment in parents of children with cancer at home in combination with parents' pain management practices. Methods used in this research are unique to investigations of parents' management of children's cancer pain at home and take a holistic approach to understanding this area. Using mixed methods, this research has confirmed that complex phenomena such as parents' management of children's cancer pain at home can be investigated within one research study. By investigating different aspects of this phenomena in a single study, relationships between aspects of the phenomena can be observed and inferences derived which would not have been achieved had a single method of data collection been used. This research has shown that complex phenomena, such as parents' management of children's cancer pain at home, can be investigated in a single study using integration of both quantitative and qualitative methods. This has implications for future researchers who may choose to base their studies on the methodology of this research when aiming to investigate complex phenomena. Use of the TPB to understand the findings is unique as no other research investigations of parents' management of children's cancer pain has utilised this theoretical framework.

### ***9.3.7 Contributions to understanding of the Theory of Planned Behaviour***

Research in this thesis makes a new contribution to understanding of TPB. Findings suggest that whilst attitudes and PBC can be used to understand behaviour for parents of children with cancer, subjective norms are not evident. Subjective norms may not be applicable in situations where individuals do not feel "normal". This includes parents of children with cancer and may include parents of children with other chronic conditions. This gap in theoretical understanding may be filled by Maslow's hierarchy of needs which suggests parents have not yet achieved basic goals for their child and therefore do not attend higher-level social needs such as subjective norms.

## **9.4 Implications**

### ***9.4.1 Implications for clinical practice***

This research found that whilst children with cancer are frequently not in pain at home, most experience episodes of clinically significant pain suggesting children's cancer pain at home may be best described with heterogeneous pain trajectories. However, neither this research nor the wider literature has ascertained a way of predicting which children will be on which pain trajectories using clinically available information. This has implications for practice where the absence of pain during hospitalisation or prior to discharge should not result in pain management interventions

being considered optional. Potential for children with cancer to experience pain at home should be considered as a matter of routine, and targeted with appropriate interventions.

Descriptions of how parents assess their child's pain illustrate how several sources of information can be used simultaneously in a bundled approach to effectively ascertain how much pain a child is in. In clinical settings, HCPs may find this a useful strategy to increase the efficacy of their own pain assessment and subsequent pain management. Attending to context, self-report, behaviours, activities, child and family preferences, emotional assessment, and risks of pharmacological or non-pharmacological interventions may improve pain assessment. This research not only provides support for this as a model of pain assessment but also delineates how this may manifest in children's cancer pain. Healthcare professionals can support families by passing on knowledge of how several elements of pain assessment can be combined in bundled approaches and the way in which each element may manifest for children's cancer pain at home. This can accelerate parents' progress through the learning phase of their pain management role.

Parents perceived that some children would not report pain due to not wanting pharmacological interventions, not wanting to go to hospital, or not wanting to think about pain. These constitute "display rules" and show how children may not accurately report their own pain. Healthcare professionals will benefit from an awareness of display rules which may be a barrier to self-report in a clinical setting. Healthcare professionals can also pass on knowledge of display rules when preparing parents for their pain management role at home.

Practical barriers to pharmacological interventions meant parents in this study felt they had an "empty toolbox". Parents were afraid of administering paracetamol for fear that its antipyretic properties could mask important signs of a fever. However, oncology protocols state that it is safe to administer paracetamol if the child is afebrile and otherwise well (Cheng and Tattermusch, 2014). Empowering parents to confidently administer paracetamol in line with oncology protocols will alleviate children's pain in these situations. Parents were also hesitant to administer morphine due to worries it would exacerbate or cause nausea or constipation. Oncology protocols recognise these side-effects, but recommend that constipation can be managed by the simultaneous administration of laxatives, which should be prescribed routinely, and anti-emetics, which should be prescribed for the first three days (Cheng and Tattermusch, 2014). Empowering parents to feel confident in administering morphine with laxatives and anti-emetics is likely to alleviate children's pain without causing negative side-effects. It is important that parents' concerns regarding pharmacological interventions are acknowledged, and that parents are given enough support to

administer these safely and effectively. A reduction in the pain experienced by children with cancer at home is likely to result from doing so.

A key finding of this research is that children find medications unpalatable, frequently refuse pharmacological interventions, and are often non-compliant. This finding has been corroborated elsewhere in literature (Bettle, 2015; Fortier *et al.*, 2014; Mariyana *et al.*, 2018). In clinical practice, HCPs may be able to support parents and children by providing alternative forms or strengths of pharmacological interventions which may improve palatability and in turn compliance. Nurses may also be aware of behavioural strategies to increase compliance which could help parents. This will reduce stress experienced by parents who undertake this in addition to many other new tasks (Clarke *et al.*, 2005). If children are compliant when parents attempt to administer pharmacological interventions, this may increase parents' efficacy in pain management and ultimately reduce pain experienced by children with cancer at home.

Evidence from inpatient cancer settings suggests HCPs underutilise non-pharmacological pain management interventions for children where they could be beneficial (Plummer *et al.*, 2017). Research in this thesis found parents preferred non-pharmacological interventions, the most common being cuddles, food/drink, distraction, massage, sleep/rest, and heat. The evidence for non-pharmacological interventions as effective pain management for children with cancer is growing (Section 8.5.3). Evidence suggests HCPs may hold negative attitudes toward non-pharmacological interventions (Stub *et al.*, 2018) which may require education or training (Cirik *et al.*, 2017). HCPs may hesitate to advise parents on treatments due to the low quality of evidence (Bao *et al.*, 2016; Coughtrey *et al.*, 2018; Jibb *et al.*, 2015; Running and Seright, 2012), but it is important parents' preferences for non-pharmacological interventions are acknowledged. Open discussion with parents regarding non-pharmacological interventions will ensure parents are aware of the implications of their use (Al-Qudimat *et al.*, 2011). If HCPs have open, supportive dialogue with parents regarding use of non-pharmacological interventions, children with cancer are more likely to have these techniques effectively applied at times when parents feel unable to use pharmacological interventions. Any risks associated with non-pharmacological interventions (Al-Qudimat *et al.*, 2011) will be minimised if HCPs are aware of their use. When HCPs work with parents to support use of non-pharmacological interventions, this will help reduce unnecessary child pain at home.

Compared to interventions targeting other individuals present in parental management of children's pain at home, the literature review in Chapter 3 found most success in increasing analgesic drug



administration and reducing child pain stemmed from interventions which targeted parents directly (Parker *et al.*, 2018). In addition, tailoring interventions was likely to increase their success. Applying these to the clinical setting suggests children are likely to receive more analgesic drug administrations, and experience a reduction in their pain, if HCPs target parents directly with support tailored around the child's diagnosis and treatment. In addition, parents may benefit from pain education, pain assessment tools, and HCPs attending to negative attitudes. Previous studies acknowledge the importance of nurses in preparing parents for their pain management role (Lu *et al.*, 2011; Twycross *et al.*, 2015a). Parents preparing to care for children with cancer at home face a steep learning curve and nurses are gatekeepers for parents' knowledge (Bettle, 2015). Nursing knowledge is not always effectively translated into practice, so the way in which parents are educated is important (Twycross, 2010). Tailored pain management education will increase parent confidence which has been associated with reduced child pain intensity and increased administration of analgesic drugs (Tutelman *et al.*, 2018).

#### ***9.4.2 Implications for future research***

Although most children with cancer are frequently not in pain at home, most experience episodes of clinically significant pain. A subset of children experience frequent clinically significant pain. Research suggests children may have heterogeneous pain trajectories (Buckner *et al.*, 2014; Wang *et al.*, 2017). To date, research has not been able to identify likely trajectories from demographic criteria or information readily available to HCPs in clinical practice. Research is needed to identify ways to enable HCPs to target children on severe pain trajectories and ensure parents are prepared for their pain management role.

This research found evidence of parents using a combination of several elements of pain assessment in bundled approaches. To date, these techniques have not been incorporated into an assessment tool so future researchers will need to design and validate tools which facilitate bundled approaches. This will enable researchers to use these techniques to enhance the accuracy of their choice of pain assessment measures in research. Researchers who incorporate bundled approaches to pain assessment in interventions aimed at supporting parents to manage their child's pain at home are likely to increase the efficacy of their intervention.

This research found that children frequently refuse pharmacological interventions due to finding them unpalatable and that parents hesitate to administer pharmacological interventions due to fear of distressing their child. Research is needed to identify ways in which parents can be supported in administering medication to children who may be non-compliant. Such interventions will reduce

parent stress associated with managing pain in children with cancer and reduce barriers to effective pharmacological interventions which ultimately reduce pain experienced by children with cancer at home. Future research could investigate the extent to which parents' use of paracetamol at home aligns with oncology protocols.

Participants found non-pharmacological interventions to be essential in managing their child's pain at home. Participants learned about these interventions through the internet, their own background, and other parents of children with cancer (Section 6.4.4.3). A lack of HCP involvement in advice around non-pharmacological interventions has been found internationally (Molassiotis *et al.*, 2010). This may be due to the lack of high quality evidence for such interventions (Bao *et al.*, 2016; Coughtrey *et al.*, 2018; Jibb *et al.*, 2015; Running and Seright, 2012). Future research which focuses on identifying effective, safe, non-pharmacological interventions for managing children's cancer pain at home will enable nurses to provide evidence-based education to parents of children with cancer. This will empower parents to effectively manage children's cancer pain at home when pharmacological interventions are contraindicated or refused by children and consequently reduce unnecessary pain experienced by children with cancer at home.

#### **9.4.3 Implications for policy**

Recent guidance for nurses on education for parents of children with cancer prior to discharge states that pain management education should be considered optional (Rodgers *et al.*, 2018). This research has shown that most children with cancer experience episodes of pain at home which are clinically significant, and cannot be predicted. Hospital policy must therefore reflect this and ensure healthcare professionals provide pain management education to all parents of children with cancer prior to discharge regardless of their pain manifestation in hospital.

Current supportive care protocols do not acknowledge the challenging analgesic context in which parents of children with cancer are managing their child's pain at home (Cheng and Tattermusch, 2014). There is a need to update these policies to acknowledge pain management challenges present due to the pharmacological context. Protocols need to ensure parents are empowered to administer pharmacological interventions safely despite this context. In addition, protocols need to ensure specific support is in place to enable parents to administer non-pharmacological interventions.

Several clinical commissioning groups in England have issued statements which do not support the use of non-mainstream practices or complementary and alternative medicines (Davies, 2018). Such statements advise against use of complementary and alternative therapies citing lack of evidence. The Government has advised NICE to consider alternative therapies but due to lack of research, they are largely unsupported. Research described in this thesis outlines the need for a policy shift. Firstly, there is a need for policy which encourages robust research into non-pharmacological interventions. In the meantime, a shift in policy is needed to empower professionals to advise safely on non-mainstream practices for which there are no safety concerns.

## **9.5 Dissemination strategy**

It is ethically important to ensure research with clinical implications is appropriately disseminated to respect the cost of participation (Akers *et al.*, 2009). The dissemination strategy for this research aims to provide findings to professionals, parents, participants, and the wider community. Parts of the strategy have already taken place, others are in progress, and others are planned. Table 9.1 provides an overview of how this research has been and will be disseminated to academic and clinical settings as well as to participants and experts by experience. Attendance at national and international conferences focused on nursing, pain, and cancer will provide opportunities to disseminate to researchers and clinicians in relevant fields. Harnessing Twitter and other social media outlets will continue to promote discussion to a varied audience of researchers, parents, clinicians, and wider community. Continued membership of research groups including the Pain in Child Health and Research in Child Health groups will enable further opportunities for dissemination.

Table 9.1: Dissemination of research findings

<b>Population</b>	<b>Strategy</b>	
Academic	<i>Dissemination to date</i>	
	<i>Planned dissemination</i>	
Academic	<u>Presentations:</u> Royal College of Nursing International Nursing Research Conference 2018, Birmingham Pain in Child Health 2018, Copenhagen European Oncology Nursing Society PhD Workshop 2018, Cyprus Pain in Child Health Webinar 2018 International Symposium on Pediatric Pain 2017, Kuala Lumpur LSBU Doctoral Support Group 2016 and 2017 RCN International Nursing Research Conference 2016, Oxford Research in Child Health 2016, London <u>Publications:</u> Parker, R., McKeever, S., Wiseman, T., and Twycross, A., (2017). An integrative review of interventions to support parents when managing their child's pain at home. <i>Pain Management Nursing</i> , November 2017	<u>Publications in progress:</u> Manifestation and parental assessment of children's cancer pain at home: a mixed methods study. The empty toolbox: Parents' experiences of managing their child's cancer pain at home. <u>Publications in planning:</u> Learnings resultant from use of pain diaries. Use of TPB in understanding parents' management of children's cancer pain at home. Secondary analysis of PPEP and MAQ results in collaboration with other authors who have used these scales.
	Clinical	LSBU undergraduate research module 2017 and 2018 LSBU postgraduate pain module 2017 and 2018 Children and Young People's Oncology Outreach Nurses clinical teaching 2017 Final report to stakeholders at The Royal Marsden NHS Foundation Trust Evidence based nursing blog and Twitter chat 2017
Experts by experience	Biannual newsletters to parents who assisted in research design Final report to parents who assisted in research design	
Participants	Final report to participants as requested	

## 9.6 Reflexive perspectives

Throughout design, data collection, analysis and write up phases, I<sup>6</sup> have exercised reflexivity to identify how my own beliefs, biases, and presence may have influenced this research (Berger,

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<sup>6</sup> The personal pronoun has been used in this section of the thesis to increase reflexivity.

2013; Parahoo, 2006). Keeping a reflective journal allowed me to consider ways in which I may have influenced my research (Jootun *et al.*, 2009). Data collection took place in a hospital where I was a staff nurse. This had both positive and negative implications. Positively, this meant whilst I put effort into building rapport with HCPs, patients, and families, to an extent, this already existed. Anecdotally, nurses commented that they trusted me and felt comfortable introducing me to potential participants. Nurses on the ward also suggested things which they thought could improve recruitment such as strategic locations for poster placement. Negatively, I worried that parents may feel coerced into the study because they knew me, especially as I was taking consent. I used every opportunity to ensure parents knew of their right to withdraw and always emphasised that their child's treatment would not be affected by their participation or non-participation. Although I cannot be certain, I took comfort from parents who I knew well and yet declined to participate as indicators that those who did participate, did so freely. My reflective journal records: *"Most parents say yes [to participation] but I am only getting back half the surveys I am giving out. Whilst this is frustrating, it is also reassuring. Parents feel they can opt out and are doing so. Parents may feel pressured to say yes but this doesn't mean they actually participate"*. This shows how my study design, specifically the anonymity of survey response, minimised ethical issues by providing potential participants with an easy way to opt out.

My reflective journal describes five ways in which the study design, setting, and conduct enhanced data collection process. Firstly, due to my position as a staff nurse within the hospital, nurses knew me and wanted to help me. In my reflective journal I wrote, *"The fact that I work here means the nurses trust me and know they are not putting their patients in harm's way. They want to help me. This makes a huge difference"*. Secondly, my perception was that parents believed the hospital delivered excellent care and as a result, patients and families felt they wanted to give back. Research provided a way to do this so, recruitment was enhanced. Thirdly, I believed data collection was enhanced by the nature of the condition of cancer. My reflective journal records: *"Cancer is a life-threatening condition. As a result, people feel that the hospital has 'saved their child's life'. If a child had appendicitis, I do not think parents would feel the same way even though that condition untreated would be life-threatening. Because of this perception, I think people are even more keen to give back"*. Fourthly, I worked hard at ensuring I described my research in a way that was easy to understand, interesting, and worthwhile. I developed a repertoire of communication strategies which enabled me to build rapport quickly and effectively. Finally, the study design allowed participants to choose the level of involvement which suited their commitment and interest. My reflective journal records: *"It also entices people in with a relatively easy starting point and then once their attention is caught there are other levels they can go to"*.

This strength of design means data collection was effective and from an ethical perspective, participants were provided with choice on extent of participation which is not often available in research.

I reflected on how my position as a nurse had potential to complicate my role as researcher and vice versa (Holloway and Galvin, 2016). It is possible parents withheld information during data collection which would have been gathered had I not been a nurse, but the converse is also true: parents may have disclosed information to me because I was a nurse (Mitchell and Irvine, 2008). During clinical nursing shifts, I did not discuss my research with patients and families unless I was asked directly. It was harder to limit conversations related to nursing within my research role, but maintaining transparency about my dual roles and limitations of both ensured I stayed within The Nursing and Midwifery Council Code at all times (NMC, 2015). Whilst I situated myself clearly within the etic perspective as I was not a parent of child with cancer, being a nurse meant that I was not entirely an outsider and could bring an element of emic perspective (Lambert *et al.*, 2011). My reflective journal records my thoughts regarding my personal experience of parenting: *“I am not a parent: I think this is a really important fact to state. This limits my empathy with my population. To be honest, I am not sure how exactly I am limited, and I think that will only become apparent if and when I become a parent. But it needs to be acknowledged. I am able to look at the situation with much more objectivity and less subjectivity than I would if I was a parent”*. I shared with parents a mutual experience of the situation, and often the child, albeit from different perspectives, which influenced data collection. My position as a nurse also had potential to influence analysis. My reflective journal records: *“...my clinical knowledge is helpful in providing some context for my findings and greater insight, but I need to be very careful that I am reporting only what is included in the results and not my own experience”*. I overcame this by constantly returning to data directly and being challenged by supervisors in our meetings.

## **9.7 Concluding remarks**

In 2011, Fortier and colleagues issued a call for research investigating children’s cancer pain at home. Introductory chapters of this thesis ascertained that insufficient evidence existed for interventions to be developed in this setting and proposed a convergent, mixed methods study which aimed to understand how parents of children with cancer manage their child’s cancer pain at home. This empirical research was combined with an integrative review aiming to identify interventions which supported parents managing their child’s pain at home and ascertain which aspects of interventions make them effective.

Integrated results found that most of the time children are not in pain. However, most children experienced episodes of clinically significant pain at home. Although heterogeneous pain trajectories may explain this divergence, research is currently not able to identify which children will experience pain and when, so all parents must be prepared for their pain management role. Parents learn to assess their child's pain using in-depth knowledge of their child's unique pain expression and several different information sources which constitute bundled approaches. Parents frequently under-medicate their child's pain but use a variety of non-pharmacological interventions to manage their child's pain at home. Paracetamol, ibuprofen, codeine, and morphine administration all have undesirable consequences, and children frequently find pharmacological interventions to be unpalatable. These are practical barriers to pharmacological interventions which mean non-pharmacological interventions are of vital importance in managing children's cancer pain at home. An integrative review provided a direction for the most effective targets for interventions to support parents managing children's pain at home. It also provided suggestions for characteristics of interventions, components of parents' pain management, and key features of research which are likely to increase intervention efficacy. This thesis presents research which will enable the development of interventions to support parents in managing their child's cancer pain at home.

## References

- Abraham, C., Kelly, M. P., West, R. and Michie, S. (2009) The UK national institute for health and clinical excellence public health guidance on behaviour change: A brief introduction, *Psychology, Health & Medicine*, 14 (1), pp. 1–8. DOI:10.1080/13548500802537903.
- Agnew & Pyke (1969) *The Science Games*. Prentice Hall.
- Ainsworth, M. D. S. (1990) Some considerations regarding theory and assessment relevant to attachments beyond infancy, *Attachment in the Preschool Years: Theory, Research, and Intervention*, pp. 463–488.
- Ajzen, I. (1985) From intentions to actions: A theory of planned behavior, in: Kuhl, J. and Beckmann, J. (eds.) *Action control*. Berlin, Heidelberg: Springer, pp. 11–39.
- Ajzen, I. (1991) The theory of planned behavior, *Organizational Behavior and Human Decision Processes*, 50 (2), pp. 179–211.
- Ajzen, I. (2005) *Attitudes, Personality, and Behavior*. McGraw-Hill Education (UK).
- Ajzen, I. (2006) *Constructing a theory of planned behavior questionnaire*. Available from: <http://people.umass.edu/~ajzen/pdf/tpb.measurement.pdf>
- Ajzen, I. (2011) The Theory of Planned Behaviour, in: Lange, P. A. M. V., Kruglanski, A. W., and Higgins, E. T. (eds.) *Handbook of Theories of Social Psychology: Volume Two: 2*. Los Angeles u.a.: SAGE Publications Ltd.,
- Akers, J., Aguiar-Ibáñez, R., Baba-Akbari Sari, A. and others (2009) CRD's Guidance for Undertaking Reviews in Health Care, *York: Centre for Reviews and Dissemination (CRD)*.
- Alberts, N. M., Gagnon, M. M. and Stinson, J. N. (2018) Chronic pain in survivors of childhood cancer: a developmental model of pain across the cancer trajectory, *Pain*, 159 (10), pp. 1916–1927.
- Aldiss, S., Baggott, C., Gibson, F., Mobbs, S. and Taylor, R. M. (2015) A critical review of the use of technology to provide psychosocial support for children and young people with long-term conditions, *Journal of Pediatric Nursing*, 30 (1), pp. 87–101. DOI:10.1016/j.pedn.2014.09.014.
- Allen, K. D. and Shriver, M. D. (1998) Role of parent-mediated pain behavior management strategies in biofeedback treatment of childhood migraines, *Behavior Therapy*, 29 (3), pp. 477–490. DOI:10.1016/S0005-7894(98)80044-0.



- Al-Qudimat, M. R., Rozmus, C. L. and Farhan, N. (2011) Family strategies for managing childhood cancer: using complementary and alternative medicine in Jordan, *Journal of Advanced Nursing*, 67 (3), pp. 591–597.
- American Academy of Pediatrics and American Pain Society (2001) The assessment and management of acute pain in infants, children, and adolescents, *Pediatrics*, 108 (3), pp. 793–797.
- American Cancer Society | Information and Resources about for Cancer: Breast, Colon, Lung, Prostate, Skin (2018). Available from: <https://www.cancer.org> [Accessed 22 May 2018].
- American Society of Clinical Oncology (2014) The state of cancer care in America, 2014: A report by the American Society of Clinical Oncology, *Journal of Oncology Practice*, 10 (2), pp. 119–142.
- Andrzejowski, P. and Carroll, W. (2016) Codeine in paediatrics: pharmacology, prescribing and controversies, *Archives of Disease in Childhood. Education and Practice Edition*, 101 (3), pp. 148–151. DOI:10.1136/archdischild-2014-307286.
- Anfara Jr, V. A. and Mertz, N. T. (2014) *Theoretical frameworks in qualitative research*. Thousand Oaks: Sage publications.
- Armitage, A. and Keeble-Ramsay, D. (2009) The rapid structured literature review as a research strategy., *Online Submission*, 6 (4), pp. 27–38.
- Armstrong, T., Almadrones, L. and Gilbert, M. R. (2005) Chemotherapy-induced peripheral neuropathy., in: *Oncology nursing forum*.32.,
- Arnstein, S. R. (1969) A ladder of citizen participation, *Journal of the American Institute of Planners*, 35 (4), pp. 216–224.
- Badr, L. K., Puzantian, H., Abboud, M., Abdallah, A. and Shahine, R. (2006) Assessing procedural pain in children with cancer in Beirut, Lebanon, *Journal of Pediatric Oncology Nursing*, 23 (6), pp. 311–320.
- Baggott, C., Cooper, B. A., Marina, N., Matthay, K. K. and Miaskowski, C. (2012) Symptom cluster analyses based on symptom occurrence and severity ratings among pediatric oncology patients during myelosuppressive chemotherapy, *Cancer Nursing*, 35 (1), pp. 19.
- Baggott, C., Cooper, B. A., Marina, N., Matthay, K. K. and Miaskowski, C. (2014) Symptom assessment in pediatric oncology: how should concordance between children’s and parents’ reports be evaluated?, *Cancer Nursing*, 37 (4), pp. 252–262.

- Baggott, C., Dodd, M., Kennedy, C., Marina, N., Matthay, K. K., Cooper, B. A. and Miaskowski, C. (2010) Changes in children's reports of symptom occurrence and severity during a course of myelosuppressive chemotherapy, *Journal of Pediatric Oncology Nursing*, 27 (6), pp. 307–315.
- Bagnasco, A., Pezzi, E., Rosa, F., Fornonil, L. and Sasso, L. (2012) Distraction techniques in children during venipuncture: an Italian experience, *Journal Of Preventive Medicine And Hygiene*, 53 (1), pp. 44–48.
- Bailey, L., Sun, J., Courtney, M. and Murphy, P. (2015) Improving postoperative tonsillectomy pain management in children: a double blinded randomised control trial of a patient analgesia information sheet, *International Journal Of Pediatric Otorhinolaryngology*, 79 (5), pp. 732–739. DOI:10.1016/j.ijporl.2015.03.003.
- Balouchi, A., Mahmoudirad, G., Hastings-Tolsma, M., Shorofi, S. A., Shahdadi, H. and Abdollahimohammad, A. (2018) Knowledge, attitude and use of complementary and alternative medicine among nurses: A systematic review, *Complementary Therapies in Clinical Practice*.
- Bandura, A. (1986) *Social foundations of thought and action: A social cognitive theory*. Vol. xiii. Englewood Cliffs, NJ, US: Prentice-Hall, Inc.
- Bandura, A. (2011) Social Cognitive Theory, in: Van Lange, P. A., Kruglanski, A. W., and Higgins, T. E. (eds.) *Handbook of Theories of Social Psychology*. 1st ed. Los Angeles: SAGE Publications Ltd,1,.
- Bao, Y. J., Hou, W., Kong, X. Y., Yang, L., Xia, J., Hua, B. J. and Knaggs, R. (2016) Hydromorphone for cancer pain, *The Cochrane Database of Systematic Reviews*, 10, pp. CD011108. DOI:10.1002/14651858.CD011108.pub2.
- Bao, Y., Kong, X., Yang, L., Liu, R., Shi, Z., Li, W., Hua, B. and Hou, W. (2014) Complementary and alternative medicine for cancer pain: an overview of systematic reviews, *Evidence-Based Complementary and Alternative Medicine*, 2014.
- Baumrind, D. (1967) Child care practices anteceding three patterns of preschool behavior, *Genetic Psychology Monographs*, 75 (1), pp. 43–88.
- Bazeley, P. (2009) Analysing mixed methods data, *Mixed Methods Research for Nursing and the Health Sciences*, pp. 84–118.
- Berger, R. (2013) Now I see it, now I don't: researcher's position and reflexivity in qualitative research, *Qualitative Research*, pp. 1468794112468475. DOI:10.1177/1468794112468475.
- Berger, Shuster and Von Roenn (2013) *Principles and Practice of Palliative Care and Supportive Oncology*. Fourth edition. Edinburgh; New York: LWW.

- Bettle, A. (2015) *Parent / carer involvement to manage pain in children with acute lymphoblastic leukemia* (PhD dissertation). Dalhousie University, Halifax, Nova Scotia.
- Birch, M. and Miller, T. (2000) Inviting intimacy: The interview as therapeutic opportunity, *International Journal of Social Research Methodology*, 3 (3), pp. 189–202.  
DOI:10.1080/13645570050083689.
- Boden, Z. V. R., Gibson, S., Owen, G. J. and Benson, O. (2015) Feelings and Intersubjectivity in Qualitative Suicide Research, *Qualitative Health Research*, pp. 1049732315576709.  
DOI:10.1177/1049732315576709.
- Borenstein, M., Hedges, L. V., Higgins, J. and Rothstein, H. R. (2009) When does it make sense to perform a meta-analysis?, *Introduction to Meta-Analysis*, pp. 357–364.
- Bossert, E., Van Cleve, L. and Savedra, M. (1996) Children with cancer: the pain experience away from the health care setting, *Journal of Pediatric Oncology Nursing*, 13 (3), pp. 109- 120 12p.
- Bowlby, J. (2005) *A secure base*. 1st ed. London: Routledge/Taylor & Francis Group.
- Bowman, K. G. (2007) A research synthesis overview, *Nursing Science Quarterly*, 20 (2), pp. 171–176. DOI:10.1177/0894318407299575.
- Braun, V. and Clarke, V. (2006) Using thematic analysis in psychology, *Qualitative Research in Psychology*, 3 (2), pp. 77–101. DOI:<http://dx.doi.org/10.1191/1478088706qp063oa>.
- Braunack-Mayer, A. (2002) The ethics of participating in research, [http://www.mja.com.au/public/issues/177\\_09\\_041102/Bra10500\\_fm.html](http://www.mja.com.au/public/issues/177_09_041102/Bra10500_fm.html). Available from: <https://digital.library.adelaide.edu.au/dspace/handle/2440/32634> [Accessed 22 October 2015].
- Brett, J., Staniszewska, S., Mockford, C., Herron-Marx, S., Hughes, J., Tysall, C. and Suleman, R. (2014) Mapping the impact of patient and public involvement on health and social care research: a systematic review, *Health Expectations*, 17 (5), pp. 637–650.
- British Pain Society (2010) Cancer Pain Management. London, England. Available from: [https://www.britishpainsociety.org/static/uploads/resources/files/book\\_cancer\\_pain.pdf](https://www.britishpainsociety.org/static/uploads/resources/files/book_cancer_pain.pdf) [Accessed 28 January 2016].
- Brown, L. D. (2003) Comparing Health Systems in Four Countries: Lessons for the United States, *American Journal of Public Health*, 93 (1), pp. 52–56.
- Bryman, A. (2006) Integrating quantitative and qualitative research: how is it done?, *Qualitative Research*, 6 (1), pp. 97–113. DOI:10.1177/1468794106058877.

- Buckland, S. (2007) *Public information pack (PIP): how to get involved in NHS, public health and social care research*. Eastleigh, Hampshire: Involve.
- Buckner, T. W., Wang, J., DeWalt, D. A., Jacobs, S., Reeve, B. B. and Hinds, P. S. (2014) Patterns of symptoms and functional impairments in children with cancer, *Pediatric Blood & Cancer*, 61 (7), pp. 1282–1288.
- Burke, N. N., Finn, D. P., McGuire, B. E. and Roche, M. (2017) Psychological stress in early life as a predisposing factor for the development of chronic pain: Clinical and preclinical evidence and neurobiological mechanisms, *Journal of Neuroscience Research*, 95 (6), pp. 1257–1270.
- Byrne, M. W., Evan, E., Goshin, L. S., Erlich, M. D., Kim, J. H. J., Saroyan, J. M. and Zeltzer, L. K. (2011) Parent self-efficacy for managing pain in seriously ill children and adolescents nearing end of life, *Palliative & Supportive Care*, 9 (2), pp. 137–147. DOI:10.1017/S1478951511000010.
- Caldwell, K., Henshaw, L. and Taylor, G. (2011) Developing a framework for critiquing health research: An early evaluation, *Nurse Education Today*, 31 (8), pp. e1–e7. DOI:10.1016/j.nedt.2010.11.025.
- Campbell, M. and Edwards, R. (2012) Ethnic differences in pain and pain management, *Pain Management*, 2 (3), pp. 219–230. DOI:10.2217/pmt.12.7.
- Campbell, M., Fitzpatrick, R., Haines, A. and Kinmonth, A. (2000) Framework for design and evaluation of complex interventions to improve health, *British Medical Journal*, 321 (7262), pp. 694.
- Cancer Research UK (2015a, May 13) Children’s cancer statistics, *Cancer Research UK*. Available from: <http://www.cancerresearchuk.org/health-professional/cancer-statistics/childrens-cancers> [Accessed 11 May 2018].
- Cancer Research UK (2015b, May 14) Teenagers’ and young adults’ cancers statistics, *Cancer Research UK*. Available from: <http://www.cancerresearchuk.org/health-professional/cancer-statistics/teenagers-and-young-adults-cancers> [Accessed 11 May 2018].
- Centre for Reviews and Dissemination (2008) *Undertaking systematic reviews of research on effectiveness CRD guidelines for those carrying out or commissioning reviews*. University of York. Available from: <http://www.opengrey.eu/item/display/10068/457284>
- Chambers, C., Reid, G. and McGrath, P. (1997) A randomized trial of a pain education booklet: effects on parents’ attitudes and postoperative pain management... presented at the meeting of the Canadian Psychological Association, Charlottetown, Prince Edward Island, June 1995, *Children’s Health Care*, 26 (1), pp. 1- 13 13p.

Cheng, D. and Tattermusch, S. (2014) Paediatric Haematology & Oncology: Supportive Care Protocols. Great Ormond Street Hospital for Children NHS Foundation Trust, The Royal Marsden NHS Foundation Trust & University College London Hospitals NHS Foundation Trust.

Children with Cancer UK (2018) Acute Lymphoblastic Leukaemia in Children | Children with Cancer UK, *Children with Cancer*. Available from:

<https://www.childrenwithcancer.org.uk/childhood-cancer-info/cancer-types/acute-lymphoblastic-leukaemia/> [Accessed 12 July 2018].

Chorney, J., Twycross, A., Mifflin, K. and Archibald, K. (2014) Can we improve parents' management of their children's postoperative pain at home?, *Pain Research and Management*, 19 (4), pp. e115–e123.

Chung, W. W., Agbayani, C.-J. G., Martinez, A., Le, V., Cortes, H., Har, K., Kain, Z. N. and Fortier, M. A. (2018) Improving Children's cancer pain management in the home setting: Development and formative evaluation of a web-based program for parents, *Computers in Biology and Medicine*.

Cırık, V., Efe, E., Öncel, S. and Gözüm, S. (2017) Experiences and attitudes of nurses regarding complementary health approaches used by themselves and their patients, *Journal of Transcultural Nursing*, 28 (4), pp. 381–390.

Clark, N. M. and Dodge, J. A. (1999) Exploring self-efficacy as a predictor of disease management, *Health Education & Behavior*, 26 (1), pp. 72–89.

Clark, N. M., Janz, N. K., Dodge, J. A. and Sharpe, P. A. (1992) Self-regulation of health behavior: the 'take PRIDE' program, *Health Education Quarterly*, 19 (3), pp. 341–354.

Clarke, J. N., Fletcher, P. C. and Schneider, M. A. (2005) Mothers' home health care work when their children have cancer, *Journal of Pediatric Oncology Nursing*, 22 (6), pp. 365–373.

Clarke, V., Braun, V. and Hayfield, N. (2015) Thematic analysis, in: Smith, J. A. (ed.) *Qualitative psychology: A practical guide to research methods*. Sage,.

Cohen, L. and Holiday, M. (1982) *Statistics for Social Scientists*. London: Harper and Row.

Cohen, L. L., Blount, R. L. and Panopoulos, G. (1997) Nurse coaching and cartoon distraction: An effective and practical intervention to reduce child, parent, and nurse distress during immunizations, *Journal of Pediatric Psychology*, 22 (3), pp. 355–370.

Cole, J. S., McCormick, A. C. and Gonyea, R. M. (2012) Respondent use of straight-lining as a response strategy in education survey research: Prevalence and implications, in: *Annual Meeting of the American Educational Research Association (AERA)*.

- Conner-Warren, R. L. (1996) Pain intensity and home pain management of children with sickle cell disease, *Issues in Comprehensive Pediatric Nursing*, 19 (3), pp. 183–195.
- Coombs, M., Parker, R. and de Vries, K. (2016) Can qualitative interviews have benefits for participants in end-of-life care research?, *European Journal of Palliative Care*, 23 (5), pp. 227–231.
- Cope, D. G. (2014) Methods and meanings: credibility and trustworthiness of qualitative research., in: *Oncology nursing forum*.41,.
- Corner & Wagland (2013) Quality of Life of Cancer Survivors in England : Analysis of Patients' Free Text Comments: Final Report. Southampton: University of Southampton.
- Coughtrey, A., Millington, A., Bennett, S., Christie, D., Hough, R., Su, M. T., *et al.* (2018) The effectiveness of psychosocial interventions for psychological outcomes in pediatric oncology: a systematic review, *Journal of Pain & Symptom Management*, 55 (3), pp. 1004–1017.  
DOI:10.1016/j.jpainsymman.2017.09.022.
- Coyne, I., Amory, A., Kiernan, G. and Gibson, F. (2014) Children's participation in shared decision-making: Children, adolescents, parents and healthcare professionals' perspectives and experiences, *European Journal of Oncology Nursing*, 18 (3), pp. 273–280.
- Craig, K. D. (1987) Age-related aspects of pain: pain in children:, *Pain*, 30, pp. S226.  
DOI:10.1016/0304-3959(87)91514-4.
- Craig, P., Dieppe, P., Macintyre, S., Michie, S., Nazareth, I. and Petticrew, M. (2008) Developing and evaluating complex interventions: the new Medical Research Council guidance, *Bmj*, 337, pp. a1655.
- Creswell, J. W. and Miller, D. L. (2000) Determining validity in qualitative inquiry, *Theory Into Practice*, 39 (3), pp. 124–130.
- Creswell, J. W. and Plano Clark, V. L. (2011) *Designing and Conducting Mixed Methods Research*. 2nd edition. Los Angeles: SAGE Publications, Inc.
- Darcy, L., Knutsson, S., Huus, K. and Enskar, K. (2014) The everyday life of the young child shortly after receiving a cancer diagnosis, from both children's and parent's perspectives, *Cancer Nursing*, 37 (6), pp. 445–456.
- Davies, S. (2018) *Position statement on the prescribing of complementary and alternative medicines*. SW London CCGs. Available from:  
<http://www.swlmcg.nhs.uk/Policies/Position%20Statements/SWL%20Position%20statement%20complementary%20and%20alternative%20medicine%20FINAL%20V2%20January%202018.pdf>  
221

Davis, M. P. and Walsh, D. (2004) Epidemiology of cancer pain and factors influencing poor pain control, *The American Journal of Hospice & Palliative Care*, 21 (2), pp. 137–142.

de Wit, R., van Dam, F., Hanneman, M., Zandbelt, L., van Buuren, A., van der Heijden, K., Leenhouts, G., Loonstra, S. and Abu-Saad, H. H. (1999) Evaluation of the use of a pain diary in chronic cancer pain patients at home, *Pain*, 79 (1), pp. 89–99.

Department of Health (2011) *Improving Outcomes: A Strategy for Cancer*. London, England.

Available from:

[https://www.gov.uk/government/uploads/system/uploads/attachment\\_data/file/213785/dh\\_123394.pdf](https://www.gov.uk/government/uploads/system/uploads/attachment_data/file/213785/dh_123394.pdf) [Accessed 28 January 2016].

Department of Health (2013) Codeine for analgesia: restricted use in children because of reports of morphine toxicity - GOV.UK. Available from: <https://www.gov.uk/drug-safety-update/codeine-for-analgesia-restricted-use-in-children-because-of-reports-of-morphine-toxicity> [Accessed 16 August 2017].

Department of Health (2015) 2010 to 2015 government policy: cancer research and treatment -

GOV.UK. Available from: <https://www.gov.uk/government/publications/2010-to-2015-government-policy-cancer-research-and-treatment/2010-to-2015-government-policy-cancer-research-and-treatment#contents> [Accessed 4 August 2015].

Department of Health and Social Care (2015) *The NHS Constitution for England*, GOV.UK.

Available from: <https://www.gov.uk/government/publications/the-nhs-constitution-for-england/the-nhs-constitution-for-england> [Accessed 22 May 2018].

Dewey, J. (1929) *Experience And Nature*. George Allen And Unwin, Limited. Available from:

<http://archive.org/details/experienceandnat029343mbp>

Dewing, J. (2007) Participatory research A method for process consent with persons who have dementia, *Dementia*, 6 (1), pp. 11–25. DOI:10.1177/1471301207075625.

Dickson, V. V., Lee, C. S. and Riegel, B. (2011) How do cognitive function and knowledge affect heart failure self-care?, *Journal of Mixed Methods Research*, 5 (2), pp. 167–189.

DOI:10.1177/1558689811402355.

DiClemente, R. J., Salazar, L. F. and Crosby, R. A. (2018) Value-expectancy theories, in:

DiClemente, R. J., Salazar, L. F., and Crosby, R. A. (eds.) *Health Behavior Theory For Public Health*. 2nd Revised edition edition. Burlington, Massachusetts: Jones and Bartlett Publishers, Inc.,

- Diorio, C., Lam, C. G., Ladas, E. J., Njuguna, F., Afungchwi, G. M., Taromina, K. and Marjerrison, S. (2016) Global use of traditional and complementary medicine in childhood cancer: A systematic review, *Journal of Global Oncology*, 3 (6), pp. 791–800.
- Dupuis, L. L., Lu, X., Mitchell, H.-R., Sung, L., Devidas, M., Mattano, L. A., *et al.* (2016) Anxiety, pain, and nausea during the treatment of standard-risk childhood acute lymphoblastic leukemia: A prospective, longitudinal study from the Children’s Oncology Group, *Cancer (0008543X)*, 122 (7), pp. 1116–1125. DOI:10.1002/cncr.29876.
- Dures, E., Rumsey, N., Morris, M. and Gleeson, K. (2011) Mixed methods in health psychology: Theoretical and practical considerations of the third paradigm, *Journal of Health Psychology*, 16 (2), pp. 332–341.
- Dwan, K., Gamble, C., Williamson, P. R. and Kirkham, J. J. (2013) Systematic review of the empirical evidence of study publication bias and outcome reporting bias—an updated review, *PloS One*, 8 (7), pp. e66844.
- Edwards, H. E., Nash, R. E., Najman, J. M., Yates, P. M., Fentiman, B. J., Dewar, A., Walsh, A. M., McDowell, J. K. and Skerman, H. M. (2001) Determinants of nurses’ intention to administer opioids for pain relief, *Nursing & Health Sciences*, 3 (3), pp. 149–159.
- Edwards, H., Walsh, A., Courtney, M., Monaghan, S., Wilson, J. and Young, J. (2007) Promoting evidence-based childhood fever management through a peer education programme based on the theory of planned behaviour, *Journal of Clinical Nursing*, 16 (10), pp. 1966–1979. DOI:10.1111/j.1365-2702.2007.01767.x.
- Elster, J. (1989) Social norms and economic theory, *The Journal of Economic Perspectives*, pp. 99–117.
- Etikan, I., Musa, S. A. and Alkassim, R. S. (2016) Comparison of convenience sampling and purposive sampling, *American Journal of Theoretical and Applied Statistics*, 5 (1), pp. 1–4.
- Family Caregiver Alliance National Center on Caregiving (2015) Selected caregiver statistics. Available from: [http://www.caregiver.org/caregiver/jsp/content\\_node.jsp?nodeid5439](http://www.caregiver.org/caregiver/jsp/content_node.jsp?nodeid5439) [Accessed 9 December 2017].
- Feilzer, M. Y. (2010) Doing mixed methods research pragmatically: implications for the rediscovery of pragmatism as a research paradigm, *Journal of Mixed Methods Research*, 4 (1), pp. 6–16. DOI:10.1177/1558689809349691.
- Ferrell, B., Rhiner, M., Shapiro, B. and Dierkes, M. (1994a) The experience of pediatric cancer pain, Part I: Impact of pain on the family, *Journal of Pediatric Nursing*, 9 (6), pp. 368- 379 12p.



- Ferrell, B., Rhiner, M., Shapiro, B. and Strause, L. (1994b) The family experience of cancer pain management in children, *Cancer Practice*, 2 (6), pp. 441- 446 6p.
- Fetters, M. D., Curry, L. A. and Creswell, J. W. (2013) Achieving integration in mixed methods designs: principles and practices, *Health Services Research*, 48 (6pt2), pp. 2134–2156.
- Fine, P. G. (2003) Maximizing benefits and minimizing risks in palliative care research that involves patients near the end of life, *Journal of Pain and Symptom Management*, 25 (4), pp. S53–S62. DOI:10.1016/S0885-3924(03)00056-3.
- Finley, G. A., Kristjánsdóttir, Ó. and Forgeron, P. A. (2009) Cultural influences on the assessment of children’s pain, *Pain Research and Management*, 14 (1), pp. 33–37.
- Firestone, W. A. (1993) Alternative arguments for generalizing from data as applied to qualitative research, *Educational Researcher*, 22 (4), pp. 16–23.
- Fishbein, M. (1979) A theory of reasoned action: Some applications and implications.
- Flury, M., Caflisch, U., Ullmann-Bremi, A. and Spichiger, E. (2011) Experiences of parents with caring for their child after a cancer diagnosis, *Journal of Pediatric Oncology Nursing*, 28 (3), pp. 143–153.
- Forgeron, P., Finlay, G. and Arnaout, M. (2006) Pediatric pain prevalence and parents’ attitudes at a cancer hospital in Jordan, *Journal of Pain & Symptom Management*, 31 (5), pp. 440- 448 9p.
- Fortier, M. A., Chung, W. W., Martinez, A., Gago-Masague, S. and Sender, L. (2016) Pain buddy: A novel use of m-health in the management of children’s cancer pain, *Computers in Biology and Medicine*, 76, pp. 202–214. DOI:10.1016/j.combiomed.2016.07.012.
- Fortier, M. A., MacLaren, J. E., Martin, S. R., Perret-Karimi, D. and Kain, Z. N. (2009) Pediatric Pain After Ambulatory Surgery: Where’s the Medication?, *Pediatrics*, 124 (4), pp. e588–e595. DOI:10.1542/peds.2008-3529.
- Fortier, M. A., Martin, S. R., Kain, D. I. and Tan, E. T. (2011a) Parental attitudes regarding analgesic use for children: differences in ethnicity and language, *Journal Of Pediatric Surgery*, 46 (11), pp. 2140–2145. DOI:10.1016/j.jpedsurg.2011.06.021.
- Fortier, M. A., Sender, L. S. and Kain, Z. N. (2011b) Management of pediatric oncology pain in the home setting: the next frontier, *Journal of Pediatric Hematology/Oncology*, 33 (4), pp. 249–250. DOI:10.1097/MPH.0b013e318217b054.

- Fortier, M. A., Wahi, A., Bruce, C., Maurer, E. L. and Stevenson, R. (2014) Pain management at home in children with cancer: A daily diary study, *Pediatric Blood & Cancer*, 61 (6), pp. 1029–1033. DOI:10.1002/pbc.24907.
- Fortier, M. A., Wahi, A., Maurer, E. L., Tan, E. T., Sender, L. S. and Kain, Z. N. (2012) Attitudes regarding analgesic use and pain expression in parents of children with cancer, *Journal of Pediatric Hematology/Oncology*, 34 (4), pp. 257–262. DOI:10.1097/MPH.0b013e318241fd07.
- Forward, S. P., Brown, T. L. and McGrath, P. J. (1996) Mothers' attitudes and behavior toward medicating children's pain, *Pain*, 67 (2–3), pp. 469–474.
- Franck, L. S., Allen, A. and Oulton, K. (2007) Making pain assessment more accessible to children and parents: Can greater involvement improve the quality of care?, *The Clinical Journal of Pain*, 23 (4), pp. 331–338. DOI:10.1097/AJP.0b013e318032456f.
- Frayne, S. M., Burns, R. B., Hardt, E. J., Rosen, A. K. and Moskowitz, M. A. (1996) The exclusion of non-English-speaking persons from research, *Journal of General Internal Medicine*, 11 (1), pp. 39–43.
- Friedrichsdorf, S. J. (2010) Pain management in children with advanced cancer and during end-of-life care, *Pediatric Hematology and Oncology*, 27 (4), pp. 257–261.
- Friedrichsdorf, S. J., Finney, D., Bergin, M., Stevens, M. and Collins, J. J. (2007) Breakthrough pain in children with cancer, *Journal of Pain and Symptom Management*, 34 (2), pp. 209–216.
- Friedrichsdorf, S. J. and Kang, T. I. (2007) The management of pain in children with life-limiting illnesses, *Pediatric Clinics Of North America*, 54 (5), pp. 645.
- Gaffney, A. and Dunne, E. A. (1986) Developmental aspects of children's definitions of pain, *Pain*, 26 (1), pp. 105–117. DOI:10.1016/0304-3959(86)90177-6.
- Gaffney, A. and Dunne, E. A. (1987) Children's understanding of the causality of pain, *PAIN®*, 29 (1), pp. 91–104. DOI:10.1016/0304-3959(87)90182-5.
- Ganann, R., Ciliska, D. and Thomas, H. (2010) Expediting systematic reviews: methods and implications of rapid reviews, *Implementation Science*, 5, pp. 56. DOI:10.1186/1748-5908-5-56.
- Gedaly-Duff, V., Lee, K. A., Nail, L., Nicholson, H. S. and Johnson, K. P. (2006) Pain, sleep disturbance, and fatigue in children with leukemia and their parents: a pilot study, *Oncology Nursing Forum*, 33 (3), pp. 641–646. DOI:10.1188/06.ONF.641-646.

- Geddes, D. M., Dones, L., Hill, E., Law, K., Harper, P. G., Spiro, S. G., Tobias, J. S. and Souhami, R. L. (1990) Quality of life during chemotherapy for small cell lung cancer: assessment and use of a daily diary card in a randomized trial, *European Journal of Cancer*, 26 (4), pp. 484–492.
- Gerrard, R., Campbell, J., Minton, O., Moback, B., Skinner, C., McGowan, C. and Stone, P. C. (2011) Achieving the preferred place of care for hospitalized patients at the end of life, *Palliative Medicine*, 25 (4), pp. 333–336. DOI:10.1177/0269216310387459.
- Gibson, F., Aldiss, S., Horstman, M., Kumpunen, S. and Richardson, A. (2010) Children and young people’s experiences of cancer care: a qualitative research study using participatory methods, *International Journal of Nursing Studies*, 47 (11), pp. 1397–1407.
- Gorji, A. H., Taebei, M., Ranjbar, M., Hatkehlouei, M. B. and Goudarzian, A. H. (2017) Effect of distraction technique and hypnosis in pain of bone marrow aspiration in children: a narrative review, *International Journal of Pediatrics-Mashhad*, 5 (3), pp. 4521–4531. DOI:10.22038/ijp.2016.20976.1754.
- Graneheim, U. H., Lindgren, B.-M. and Lundman, B. (2017) Methodological challenges in qualitative content analysis: A discussion paper, *Nurse Education Today*, 56, pp. 29–34.
- Graneheim, U. H. and Lundman, B. (2004) Qualitative content analysis in nursing research: concepts, procedures and measures to achieve trustworthiness, *Nurse Education Today*, 24 (2), pp. 105–112. DOI:10.1016/j.nedt.2003.10.001.
- Green, J. and Thorogood, N. (2013) *Qualitative Methods for Health Research*. SAGE.
- Greene, J. C. (2008) Is mixed methods social inquiry a distinctive methodology?, *Journal of Mixed Methods Research*, 2 (1), pp. 7–22.
- Greene, J. C. and Caracelli, V. J. (1997) Defining and describing the paradigm issue in mixed-method evaluation, *New Directions for Evaluation*, 1997 (74), pp. 5–17. DOI:10.1002/ev.1068.
- Greene, J. C., Caracelli, V. J. and Graham, W. F. (1989) Toward a Conceptual Framework for Mixed-Method Evaluation Designs, *Educational Evaluation and Policy Analysis*, 11 (3), pp. 255–274. DOI:10.3102/01623737011003255.
- Guba, D. E. G. (1990) The alternative paradigm dialog, in: Guba, D. E. G. (ed.) *The Paradigm Dialog*. Newbury Park, Calif.: Sage Publications, Inc.,
- Guetterman, T. C., Fetters, M. D. and Creswell, J. W. (2015) Integrating quantitative and qualitative results in health science mixed methods research through joint displays, *The Annals of Family Medicine*, 13 (6), pp. 554–561.

- Gupta, R. (2018) Cancer Pain, in: *Multiple Choice Questions in Pain Management*. Springer, pp. 115–124.
- Haidich, A. B. (2010) Meta-analysis in medical research, *Hippokratia*, 14 (Suppl 1), pp. 29–37.
- Ham, M., Jeger, M. and Ivković, A. F. (2015) The role of subjective norms in forming the intention to purchase green food, *Economic Research-Ekonomska Istraživanja*, 28 (1), pp. 738–748.  
DOI:10.1080/1331677X.2015.1083875.
- Hanmod, S. S. and Gera, R. (2016) Oncologic pain in pediatrics, *Journal of Pain Management*, 9 (2), pp. 165–175.
- Haraldstad, K., Sørum, R., Eide, H., Natvig, G. K. and Helseth, S. (2011) Pain in children and adolescents: prevalence, impact on daily life, and parents' perception, a school survey, *Scandinavian Journal of Caring Sciences*, 25 (1), pp. 27–36. DOI:10.1111/j.1471-6712.2010.00785.x.
- Hardeman, W., Johnston, M., Johnston, D., Bonetti, D., Wareham, N. and Kinmonth, A. L. (2002) Application of the theory of planned behaviour in behaviour change interventions: A systematic review, *Psychology and Health*, 17 (2), pp. 123–158.
- Harris, M. and Taylor, G. (2008) *Medical Statistics Made Easy, second edition*. 2Rev Ed edition. Bloxham, Oxfordshire: Scion Publishing Ltd.
- Health Research Authority (2016) *Public involvement in research and research ethics committee review*. Available from: <http://www.hra.nhs.uk/documents/2016/05/hra-involve-updated-statement-2016.pdf>
- Health Research Authority (2018) General Principles - Consent and Participant information sheet preparation guidance. Available from: <http://www.hra-decisiontools.org.uk/consent/principles-general.html> [Accessed 15 June 2018].
- Heavey, E. (2014) *Statistics for nursing: A practical approach*. Jones & Bartlett Publishers.
- Hechler, T., Chalkiadis, G. A., Hasan, C., Kosfelder, J., Meyerhoff, U., Vocks, S. and Zernikow, B. (2009) Sex differences in pain intensity in adolescents suffering from cancer: differences in pain memories?, *The Journal of Pain*, 10 (6), pp. 586–593.
- Hedén, L., Pöder, U., von Essen, L. and Ljungman, G. (2013) Parents' perceptions of their child's symptom burden during and after cancer treatment, *Journal of Pain and Symptom Management*, 46 (3), pp. 366–375.

- Hedström, M., Haglund, K., Skolin, I. and Von Essen, L. (2003) Distressing events for children and adolescents with cancer: Child, parent, an nurse perceptions, *Journal of Pediatric Oncology Nursing*, 20 (3), pp. 120–132.
- Hegarty, M., Calder, A., Davies, K., Shave, M., Christiansen, E., Meyer, T. and von Ungern-Sternberg, B. S. (2013) Does take-home analgesia improve postoperative pain after elective day case surgery? A comparison of hospital vs parent-supplied analgesia, *Paediatric Anaesthesia*, 23 (5), pp. 385–389. DOI:10.1111/pan.12077.
- Helgadóttir, H. L. and Wilson, M. E. (2014) A randomized controlled trial of the effectiveness of educating parents about distraction to decrease postoperative pain in children at home after tonsillectomy, *Pain Management Nursing*, 15 (3), pp. 632- 640 9p. DOI:10.1016/j.pmn.2013.07.001.
- Hellsten, M. B. (2000) All the king's horses and all the king's men: Pain management from hospital to home, *Journal of Pediatric Oncology Nursing*, 17 (3), pp. 149–159.
- Hochbaum, G., Rosenstock, I. and Kegels, S. (1952) Health belief model, *United States Public Health Service*. Available from: [http://www.infosihat.gov.my/infosihat/artikelHP/bahanrujukan/HE\\_DAN\\_TEORI/DOC/Health%20Belief%20Model.doc](http://www.infosihat.gov.my/infosihat/artikelHP/bahanrujukan/HE_DAN_TEORI/DOC/Health%20Belief%20Model.doc) [Accessed 20 January 2016].
- Holloway, I. and Galvin, K. (2016) *Qualitative research in nursing and healthcare*. John Wiley & Sons.
- Holloway, I. and Wheeler, S. (2013) *Qualitative research in nursing and healthcare*. John Wiley & Sons. Available from: <https://books.google.co.uk/books?hl=en&lr=&id=CmV32bhRaowC&oi=fnd&pg=PR11&dq=qualitative+research+in+nursing+holloway+and+wheeler+2002&ots=7bq65cE-RZ&sig=FvaZ43ygIs2ycX5qqK4fCgTGH0g>
- Howe, K. R. (1988) Against the quantitative-qualitative incompatibility thesis or dogmas die hard, *Educational Researcher*, 17 (8), pp. 10–16. DOI:10.3102/0013189X017008010.
- Howitt, D. and Cramer, D. (2010) Chapter 21: Thematic analysis, in: Howitt, D. (ed.) *Introduction to Research Methods in Psychology*. 2nd edition. Harlow: Prentice Hall,.
- Hsieh, H.-F. and Shannon, S. E. (2005) Three approaches to qualitative content analysis, *Qualitative Health Research*, 15 (9), pp. 1277–1288.

Huby, K., Swallow, V., Smith, T. and Carolan, I. (2017) Children and young people's views on access to a web-based application to support personal management of long-term conditions: a qualitative study, *Child: Care, Health and Development*, 43 (1), pp. 126–132.

Involve (2013) *Exploring the impact of public involvement on the quality of research: examples*. Involve Eastleigh, UK.

Irvine, L. (2017) Childhood cancer statistics – what can we learn from new data? - Public health matters. Available from: <https://publichealthmatters.blog.gov.uk/2017/02/01/childhood-cancer-statistics-what-can-we-learn-from-new-data/> [Accessed 8 September 2017].

Janke, J. R. (1994) Development of the Breast-Feeding Attrition Prediction Tool, *Nursing Research*, 43 (2), pp. 100–104.

Jekauc, D., Völkle, M., Wagner, M. O., Mess, F., Reiner, M. and Renner, B. (2015) Prediction of attendance at fitness center: a comparison between the theory of planned behavior, the social cognitive theory, and the physical activity maintenance theory, *Frontiers in Psychology*, 6. DOI:10.3389/fpsyg.2015.00121.

Jibb, L. A., Croal, L., Wang, J., Yuan, C., Foster, J., Cheung, V., Gladstone, B. M. and Stinson, J. N. (2018) Children's experiences of cancer care: a systematic review and thematic synthesis of qualitative studies., in: *Oncology nursing forum*.45, pp. 527–544.

Jibb, L. A., Nathan, P. C., Stevens, B. J., Seto, E., Cafazzo, J. A., Stephens, N., Yohannes, L. and Stinson, J. N. (2015) Psychological and physical interventions for the management of cancer-related pain in pediatric and young adult patients: an integrative review., in: *Oncology nursing forum*.42,.

Johnson, B. and Turner, L. (2003) Data collection strategies in mixed methods research, in: Tashakkori, A. and Teddlie, C. (eds.) *Handbook of Mixed Methods in Social & Behavioral Research*. SAGE,.

Johnson, R. B. and Onwuegbuzie, A. J. (2004) Mixed methods research: a research paradigm whose time has come, *Educational Researcher*, 33 (7), pp. 14–26. DOI:10.3102/0013189X033007014.

Jones, M. and Rattray, J. (2010) Questionnaire design, in: *The research process in nursing*. John Wiley & Sons,.

Jootun, D., McGhee, G. and Marland, G. R. (2009) Reflexivity: promoting rigour in qualitative research, *Nursing Standard*, 23 (23), pp. 42.

- Kankkunen, P., Pietilä, A.-M. and Vehviläinen-Julkunen, K. (2004) Families' and children's postoperative pain--literature review, *Journal of Pediatric Nursing*, 19 (2), pp. 133–139.
- Kankkunen, P., Vehviläinen-Julkunen, K., Pietilä, A., Kokki, H., Grey, M., Kain, Z. and Zisk, Y. (2008) A tale of two countries: Comparison of the perceptions of analgesics among Finnish and American parents, *Pain Management Nursing*, 9 (3), pp. 113–119.  
DOI:10.1016/j.pmn.2007.12.003.
- Kankkunen, P., Vehviläinen-Julkunen, K., Pietilä, A., Kokki, H. and Halonen, P. (2003) Parents' perceptions and use of analgesics at home after children's day surgery, *Paediatric Anaesthesia*, 13 (2), pp. 132–140.
- Kankkunen, P., Vehviläinen-Julkunen, K., Pietilä, A.-M., Korhonen, A., Nyssönen, S., Lehikoinen, N.-M. and Kokki, H. (2009) Promoting children's pharmacological postoperative pain alleviation at home, *Pediatric Nursing*, 35 (5), pp. 298–303; quiz 304–305.
- Kars, M. C., Duijnste, M. S. H., Pool, A., van Delden, J. J. M. and Grypdonck, M. H. F. (2008) Being there: parenting the child with acute lymphoblastic leukaemia, *Journal of Clinical Nursing*, 17 (12), pp. 1553–1562. DOI:10.1111/j.1365-2702.2007.02235.x.
- Kazak, A. E. and Noll, R. B. (2015) The integration of psychology in pediatric oncology research and practice: collaboration to improve care and outcomes for children and families, *The American Psychologist*, 70 (2), pp. 146–158. DOI:10.1037/a0035695.
- Kearney, N., McCann, L., Norrie, J., Taylor, L., Gray, P., McGee-Lennon, M., Sage, M., Miller, M. and Maguire, R. (2009) Evaluation of a mobile phone-based, advanced symptom management system (ASyMS\copyright) in the management of chemotherapy-related toxicity, *Supportive Care in Cancer*, 17 (4), pp. 437–444.
- Klitbo, D. M., Nielsen, R., Illum, N. O., Wehner, P. S. and Carlsen, N. (2011) Symptoms and time to diagnosis in children with brain tumours, *Danish Medical Bulletin*, 58 (7), pp. A4285.
- Kritsonis, A. (2005) Comparison of change theories, *International Journal of Scholarly Academic Intellectual Diversity*, 8 (1), pp. 1–7.
- Krueger, C. (2008) *Focus Groups: A Practical Guide for Applied Research*. 4th edition. Los Angeles: SAGE Publications, Inc.
- Kuper, A., Lingard, L. and Levinson, W. (2008) Critically appraising qualitative research, *Bmj*, 337 (07 (3)), pp. a1035–a1035.
- Lambert, V., Glacken, M. and McCarron, M. (2011) Employing an ethnographic approach: key characteristics, *Nurse Researcher (through 2013)*, 19 (1), pp. 17.

- LaMontagne, L. L., Wells, N., Hepworth, J. T., Johnson, B. D. and Manes, R. (1999) Parent coping and child distress behaviors during invasive procedures for childhood cancer, *Journal of Pediatric Oncology Nursing*, 16 (1), pp. 3–12.
- Latter, S., Hopkinson, J. B., Lowson, E., Hughes, J. A., Hughes, J., Duke, S., *et al.* (2018) Supporting carers to manage pain medication in cancer patients at the end of life: A feasibility trial, *Palliative Medicine*, 32 (1), pp. 246–256.
- Lautenbacher, S., Peters, J. H., Heesen, M., Scheel, J. and Kunz, M. (2017) Age changes in pain perception: a systematic-review and meta-analysis of age effects on pain and tolerance thresholds, *Neuroscience & Biobehavioral Reviews*, 75, pp. 104–113.
- Legare, F., Boivin, A., van der Weijden, T., Pakenham, C., Burgers, J., Legare, J., St-Jacques, S. and Gagnon, S. (2011) Patient and Public Involvement in Clinical Practice Guidelines: A Knowledge Synthesis of Existing Programs, *Medical Decision Making*, 31 (6), pp. E45–E74. DOI:10.1177/0272989X11424401.
- Liberati, A., Altman, D. G., Tetzlaff, J., Mulrow, C., Gøtzsche, P. C., Ioannidis, J. P., *et al.* (2009) The PRISMA statement for reporting systematic reviews and meta-analyses of studies that evaluate health care interventions: explanation and elaboration, *Annals of Internal Medicine*, 151 (4), pp. W–65.
- Ljungman, G., Gordh, T., Sörensen, S. and Kreuger, A. (1999) Pain in paediatric oncology: interviews with children, adolescents and their parents, *Acta Paediatrica*, 88 (6), pp. 623–630.
- Ljungman, G., Kreuger, A., Gordh, T. and Sörensen, S. (2006) Pain in pediatric oncology: do the experiences of children and parents differ from those of nurses and physicians?, *Uppsala Journal of Medical Sciences*, 111 (1), pp. 87–95.
- Longard, J., Twycross, A., Williams, A., Hong, P. and Chorney, J. (2016) Parents' experiences of managing their child's postoperative pain at home: an exploratory qualitative study, *Journal of Clinical Nursing*, 25 (17–18), pp. 2619–2628.
- Lovell, M. R., Luckett, T., Boyle, F. M., Phillips, J., Agar, M. and Davidson, P. M. (2014) Patient education, coaching, and self-management for cancer pain., *Journal of Clinical Oncology: Official Journal of the American Society of Clinical Oncology*.
- Lu, C.-H., Huang, C.-Y., Park, J.-H., Lin, H.-R., Lee, Y.-L. and Cheng, S.-F. (2011) Main caregivers' experiences of managing pain for children with neuroblastoma in Taiwan, *Journal of Pediatric Oncology Nursing*, 28 (6), pp. 326–335. DOI:10.1177/1043454211408104.



- Lynch, M. K., Dimmitt, R. A. and Goodin, B. R. (2018) Evidence of disturbed sleep in children with eosinophilic esophagitis and persistent epigastric pain, *Journal of Pediatric Psychology*, 43 (3), pp. 331–341. DOI:10.1093/jpepsy/jsx117.
- Madi, D. and Clinton, M. (2018) Pain and its impact on the functional ability in children treated at the children's cancer center of Lebanon, *Journal of Pediatric Nursing*.
- Marie, N., Lockett, T., Davidson, P. M., Lovell, M. and Lal, S. (2013) Optimal patient education for cancer pain: a systematic review and theory-based meta-analysis, *Supportive Care in Cancer*, 21 (12), pp. 3529–3537.
- Mariyana, R., Allenidekania, A. and Nurhaeni, N. (2018) Parents' voice in managing the pain of children with cancer during palliative care, *Indian Journal of Palliative Care*, 24 (2), pp. 156–161. DOI:10.4103/IJPC.IJPC\_198\_17.
- Marks, A. D., Keefer, P. and Saul, D. (2013) Use of intravenous acetaminophen (paracetamol) in a pediatric patient at the end of life: Case report, *Journal of Palliative Medicine*, 16 (12), pp. 1641–1643.
- Maslow, A. H. (1943) A theory of human motivation., *Psychological Review*, 50 (4), pp. 370.
- Maslow, A. and Lewis, K. J. (1987) Maslow's hierarchy of needs, *Salenger Incorporated*, 14, pp. 987.
- Mason, M. (2010) Sample Size and Saturation in PhD Studies Using Qualitative Interviews, *Forum Qualitative Sozialforschung / Forum: Qualitative Social Research*, 11 (3). Available from: <http://www.qualitative-research.net/index.php/fqs/article/view/1428> [Accessed 29 April 2016].
- McClain, B. C. and Suresh, S. (eds.) (2011) *Handbook of Pediatric Chronic Pain*. New York, NY: Springer New York. Available from: <http://link.springer.com/10.1007/978-1-4419-0350-1>
- McKenna, H., Hasson, F. and Keeney, S. (2010) Surveys, in: Gerrish, K. and Lacey, A. (eds.) *The Research Process in Nursing*. John Wiley & Sons,.
- McLaughlin, H. (2009) What's in a name: 'Client', 'Patient', 'Customer', 'Consumer', 'Expert by Experience', 'Service User'—What's next?, *The British Journal of Social Work*, 39 (6), pp. 1101–1117. DOI:10.1093/bjsw/bcm155.
- McMillan, J. H. and Schumacher, S. (2010) *Research in Education: Evidence-Based Inquiry*, MyEducationLab Series., *Pearson*.
- Melnyk, B. M., Alpert-Gillis, L., Feinstein, N. F., Crean, H. F., Johnson, J., Fairbanks, E., *et al.* (2004) Creating opportunities for parent empowerment: program effects on the mental

health/coping outcomes of critically ill young children and their mothers, *Pediatrics*, 113 (6), pp. e597–e607.

Melnyk, B. and Morrison-Beedy, D. (2012) *Intervention Research: Designing, Conducting, Analyzing, and Funding*. Springer Publishing Company. Available from: [https://books.google.com/books?hl=en&lr=&id=uGF5h8f7CWMC&oi=fnd&pg=PR1&dq=cost+of+funding+intervention+research&ots=0cjff0mjOF&sig=\\_R19FURxWqcpG7TKed9Mw8YXeTs](https://books.google.com/books?hl=en&lr=&id=uGF5h8f7CWMC&oi=fnd&pg=PR1&dq=cost+of+funding+intervention+research&ots=0cjff0mjOF&sig=_R19FURxWqcpG7TKed9Mw8YXeTs)

MHRA (2013) Codeine for analgesia: restricted use in children because of reports of morphine toxicity, *GOV.UK*. Available from: <https://www.gov.uk/drug-safety-update/codeine-for-analgesia-restricted-use-in-children-because-of-reports-of-morphine-toxicity> [Accessed 26 April 2018].

Michie, S., Fixsen, D., Grimshaw, J. M. and Eccles, M. P. (2009) Specifying and reporting complex behaviour change interventions: the need for a scientific method, *Implementation Science*, 4, pp. 40. DOI:10.1186/1748-5908-4-40.

Miles, M. B. and Huberman, A. M. (1994) *Qualitative data analysis: An expanded sourcebook*. Sage.

Miller, E., Jacob, E. and Hockenberry, M. J. (2011) Nausea, pain, fatigue, and multiple symptoms in hospitalized children with cancer, *Oncology Nursing Forum*, 38 (5), pp. E382-393. DOI:10.1188/11.ONF.E382-E393.

Miller, M. M., Donald, D. V. and Hagemann, T. M. (2012) Prevention and Treatment of Oral Mucositis in Children with Cancer, *The Journal of Pediatric Pharmacology and Therapeutics*, 17 (4), pp. 340–350. DOI:10.5863/1551-6776-17.4.340.

Mitchell, W. and Irvine, A. (2008) I'm okay, you're okay?: Reflections on the well-being and ethical requirements of researchers and research participants in conducting qualitative fieldwork interviews, *International Journal of Qualitative Methods*, 7 (4), pp. 31–44.

Molassiotis, A., Wengström, Y. and Kearney, N. (2010) Symptom cluster patterns during the first year after diagnosis with cancer, *Journal of Pain and Symptom Management*, 39 (5), pp. 847–858.

Molinaro, M. L. and Fletcher, P. C. (2018) The Balancing Act: Mothers' Experiences of Providing Care to Their Children With Cancer, *Journal of Pediatric Oncology Nursing*, pp. 1043454218794667.

Mora, E., Smith, E. M. L., Donohoe, C. and Hertz, D. L. (2016) Vincristine-induced peripheral neuropathy in pediatric cancer patients, *American Journal of Cancer Research*, 6 (11), pp. 2416–2430.

- Moran-Ellis, J., Alexander, V. D., Cronin, A., Dickinson, M., Fielding, J., Slaney, J. and Thomas, H. (2006) Triangulation and integration: processes, claims and implications, *Qualitative Research*, 6 (1), pp. 45–59.
- Morren, M., van Dulmen, S., Ouwkerk, J. and Bensing, J. (2009) Compliance with momentary pain measurement using electronic diaries: A systematic review, *European Journal of Pain*, 13 (4), pp. 354–365. DOI:10.1016/j.ejpain.2008.05.010.
- Morse, J. M., Barrett, M., Mayan, M., Olson, K. and Spiers, J. (2002) Verification strategies for establishing reliability and validity in qualitative research, *International Journal of Qualitative Methods*, 1 (2), pp. 13–22.
- Morton, L. M. (2008) Encouraging participation in medical research: what strategies work?, *Journal of Clinical Epidemiology*, 61 (10), pp. 969–970. DOI:10.1016/j.jclinepi.2007.09.001.
- Myles, P. and Gin, T. (2000) *Statistical Methods for Anaesthesia and Intensive Care, 1e*. Oxford England; Boston: Butterworth-Heinemann.
- Nash, R., Edwards, H. and Nebauer, M. (1993) Effect of attitudes, subjective norms and perceived control on nurses' intention to assess patients' pain, *Journal of Advanced Nursing*, 18 (6), pp. 941–947.
- National Cancer Institute (2015) A Snapshot of Pediatric Cancers, *National Cancer Institute*. cgvArticle. Available from: <http://www.cancer.gov/research/progress/snapshots/pediatric> [Accessed 27 January 2016].
- Newington, L. and Metcalfe, A. (2014) Factors influencing recruitment to research: qualitative study of the experiences and perceptions of research teams, *BMC Medical Research Methodology*, 14, pp. 10. DOI:10.1186/1471-2288-14-10.
- NHS England (2013) 2013/14 NHS Standard Contract for Paediatric Oncology. NHS Commissioning Board. Available from: <https://www.england.nhs.uk/wp-content/uploads/2013/06/e04-paedi-oncol.pdf> [Accessed 22 January 2016].
- NHS England (2017) 2013/14 NHS Standard Contract for Paediatric Oncology. Available from: <https://www.england.nhs.uk/publication/201314-nhs-standard-contract-for-paediatric-oncology/> [Accessed 10 May 2018].
- NHS Quality Improvement Scotland (2009) The management of pain in patients with cancer. Edinburgh, Scotland. Available from: [Accessed 22 January 2016].

NICE (2005) Improving outcomes in children and young people with cancer | Guidance and guidelines | NICE. Available from: <https://www.nice.org.uk/guidance/csg7> [Accessed 22 January 2016].

NICE (2007) Behaviour change: general approaches | introduction | Guidance and guidelines | NICE. Available from: <https://www.nice.org.uk/guidance/ph6/chapter/Introduction> [Accessed 15 February 2016].

NICE (2014) Cancer services for children and young people | Guidance and guidelines | NICE. Available from: <https://www.nice.org.uk/guidance/qs55> [Accessed 22 January 2016].

Nishimura, A., Carey, J., Erwin, P. J., Tilburt, J. C., Murad, M. H. and McCormick, J. B. (2013) Improving understanding in the research informed consent process: a systematic review of 54 interventions tested in randomized control trials, *BMC Medical Ethics*, 14, pp. 28.  
DOI:10.1186/1472-6939-14-28.

NMC (2015) The Code for nurses and midwives. Available from: <https://www.nmc.org.uk/standards/code/> [Accessed 25 March 2016].

Noble, H. and Smith, J. (2015) Issues of validity and reliability in qualitative research, *Evidence-Based Nursing*, pp. ebnurs-2015-102054. DOI:10.1136/eb-2015-102054.

Noel, M., McMurtry, C. M., Chambers, C. T. and McGrath, P. J. (2010) Children's memory for painful procedures: the relationship of pain intensity, anxiety, and adult behaviours to subsequent recall, *Journal of Pediatric Psychology*, 35 (6), pp. 626–636. DOI:10.1093/jpepsy/jsp096.

NRES (2016) *NRES shared single issue ethical debate: issue paper one - time to consent*. National Patient Safety Agency. Available from: <http://www.hra.nhs.uk/documents/2013/09/issues-and-arguments-time-to-consent.pdf>

Oakes, L. (2011) Pain causes and treatment, in: Baggott, C., Kelly, K., Fochtman, D., and Foley, V. (eds.) *Nursing care of children and adolescents with cancer and blood disorders*. 4th ed. Chicago, IL: Association of Pediatric Hematology/Oncology Nurses, pp. 642–668.

Oberoi, S., Suthar, R., Bansal, D. and Marwaha, R. K. (2013) Febrile neutropenia: outline of management, *The Indian Journal of Pediatrics*, 80 (2), pp. 138–143.

O'Cathain, A. (2006) *Exploiting the potential of mixed methods studies on health services research*. University of Sheffield. Available from : <http://europepmc.org/abstract/eth/434506>

O'Cathain, A., Murphy, E. and Nicholl, J. (2010) Three techniques for integrating data in mixed methods studies, *BMJ*, 341, pp. c4587. DOI:10.1136/bmj.c4587.

O’Cathain, A. and Thomas, K. (2006) Combining qualitative and quantitative methods, in: Pope, C. and Mays, N. (eds.) *Qualitative research in health care*. Malden, MA. USA: BMJ Books, Blackwell Publishing LTD,.

Office for National Statistics (2012) 2011 Census: Key statistics for England and Wales, March 2011. Available from:  
<https://www.ons.gov.uk/peoplepopulationandcommunity/populationandmigration/populationestimates/bulletins/2011censuskeystatisticsforenglandandwales/2012-12-11#ethnic-group> [Accessed 12 September 2017].

Olson, K. and Amari, A. (2015) Self-reported pain in adolescents with leukemia or a brain tumor: A systematic review, *Cancer Nursing*, 38 (5), pp. E43–E53.

Onwuegbuzie, A. J. and Johnson, R. B. (2006) The validity issue in mixed research, *Research in the Schools*, 13 (1), pp. 48–63.

Opsal, T., Wolgemuth, J., Cross, J., Kaanta, T., Dickmann, E., Colomer, S. and Erdil-Moody, Z. (2016) “There are no known benefits...” Considering the risk/benefit ratio of qualitative research, *Qualitative Health Research*, 26 (8), pp. 1137–1150.

Owen, L., Morgan, A., Fischer, A., Ellis, S., Hoy, A. and Kelly, M. P. (2012) The cost-effectiveness of public health interventions, *Journal of Public Health*, 34 (1), pp. 37–45. DOI:10.1093/pubmed/fdr075.

Paediatric Formulary Committee (2016) *BNF for Children (BNFC) 2016-2017*. England: Pharmaceutical Press.

Palermo, T. M. (2000) Impact of recurrent and chronic pain on child and family daily functioning: a critical review of the literature., *Journal of Developmental and Behavioral Pediatrics*, 21, pp. 58–69.

Palermo, T. M., Law, E., Churchill, S. S. and Walker, A. (2012) Longitudinal course and impact of insomnia symptoms in adolescents with and without chronic pain, *The Journal of Pain*, 13 (11), pp. 1099–1106. DOI:10.1016/j.jpain.2012.08.003.

Palermo, T. M., Wilson, A. C., Lewandowski, A. S., Toliver-Sokol, M. and Murray, C. B. (2011) Behavioral and psychosocial factors associated with insomnia in adolescents with chronic pain, *Pain*, 152 (1), pp. 89–94. DOI:10.1016/j.pain.2010.09.035.

Palermo, T. M., Wilson, A. C., Peters, M., Lewandowski, A. and Somhegyi, H. (2009) Randomized controlled trial of an Internet-delivered family cognitive-behavioral therapy

intervention for children and adolescents with chronic pain, *Pain*, 146 (1–2), pp. 205–213.  
DOI:10.1016/j.pain.2009.07.034.

Paquette, J., Le May, S., Lachance Fiola, J., Villeneuve, E., Lapointe, A. and Bourgault, P. (2013) A randomized clinical trial of a nurse telephone follow-up on paediatric tonsillectomy pain management and complications, *Journal Of Advanced Nursing*, 69 (9), pp. 2054–2065.  
DOI:10.1111/jan.12072.

Parahoo (2006) *Nursing Research: Principles, Process and Issues*. 2nd edition. Basingstoke, Hampshire England; New York: Palgrave Macmillan.

Parahoo, K. (2014) *Nursing Research: Principles, Process and Issues*. 3rd Revised edition edition. Palgrave Macmillan.

Parker, R., McKeever, S., Wiseman, T. and Twycross, A. (2018) An integrative review of interventions to support parents when managing their child's pain at home, *Pain Management Nursing*, 19 (2), pp. 139–156.

Pashankar, F. D., Season, J. H., McNamara, J. and Pashankar, D. S. (2011) Acute Constipation in Children Receiving Chemotherapy for Cancer, *Journal of Pediatric Hematology/Oncology*, 33 (7), pp. e300. DOI:10.1097/MPH.0b013e31821a0795.

Penn, A., Lewis, S. P., Hunt, L. P., Shortman, R. I., Stevens, M. C., McCarter, R. L., Curran, A. L. and Sharples, P. M. (2008) Health related quality of life in the first year after diagnosis in children with brain tumours compared with matched healthy controls; a prospective longitudinal study, *European Journal of Cancer*, 44 (9), pp. 1243–1252.

Petros, S. G. (2011) Use of a mixed methods approach to investigate the support needs of older caregivers to family members affected by HIB and AIDS in South Africa, *Journal of Mixed Methods Research*. DOI:10.1177/1558689811425915.

Piaget, J., Cook, M. and Norton, W. W. (1952) *The origins of intelligence in children*. Vol. 8. International Universities Press New York. Available from:  
[http://www.bxscience.edu/ourpages/auto/2014/11/16/50007779/Piaget%20When%20Thinking%20Begins10272012\\_0000.pdf](http://www.bxscience.edu/ourpages/auto/2014/11/16/50007779/Piaget%20When%20Thinking%20Begins10272012_0000.pdf)

Plummer, K., McCarthy, M., McKenzie, I., Newall, F. and Manias, E. (2017) Pain assessment and management in paediatric oncology: a cross-sectional audit, *Journal of Clinical Nursing*, 26 (19/20), pp. 2995–3006. DOI:10.1111/jocn.13643.

Pöder, U., Ljungman, G. and von Essen, L. (2010) Parents' perceptions of their children's cancer-related symptoms during treatment: a prospective, longitudinal study, *Journal of Pain and Symptom Management*, 40 (5), pp. 661–670. DOI:10.1016/j.jpainsymman.2010.02.012.

Polit, D. F. and Beck, C. T. (2010) Generalization in quantitative and qualitative research: Myths and strategies, *International Journal of Nursing Studies*, 47 (11), pp. 1451–1458.

Popay, J., Roberts, H., Sowden, A., Petticrew, M., Arai, L., Rodgers, M., Britten, N., Roen, K. and Duffy, S. (2006) Guidance on the conduct of narrative synthesis in systematic reviews, *A Product from the ESRC Methods Programme Version*, 1. Available from:

[https://www.researchgate.net/profile/Lisa\\_Arai/publication/242311393\\_Guidance\\_on\\_the\\_conduct\\_of\\_narrative\\_synthesis\\_in\\_systematic\\_reviews\\_a\\_comparison\\_of\\_guidance-led\\_narrative\\_synthesis\\_versus\\_meta-analysis/links/5532159f0cf2f2a588ad67fd.pdf](https://www.researchgate.net/profile/Lisa_Arai/publication/242311393_Guidance_on_the_conduct_of_narrative_synthesis_in_systematic_reviews_a_comparison_of_guidance-led_narrative_synthesis_versus_meta-analysis/links/5532159f0cf2f2a588ad67fd.pdf) [Accessed 31 August 2016].

Pope, C. and Mays, N. (eds.) (2006) *Qualitative Research in Health Care Third Edition*. Revised edition. Malden, Mass.: BM.

Post-White, J. (2006) Complementary and alternative medicine in pediatric oncology, *Journal of Pediatric Oncology Nursing*, 23 (5), pp. 244–253.

Post-White, J., Fitzgerald, M., Hageness, S. and Sencer, S. F. (2009a) Complementary and alternative medicine use in children with cancer and general and specialty pediatrics, *Journal of Pediatric Oncology Nursing*, 26 (1), pp. 7–15.

Post-White, J., Fitzgerald, M., Savik, K., Hooke, M. C., Hannahan, A. B. and Sencer, S. F. (2009b) Massage therapy for children with cancer, *Journal of Pediatric Oncology Nursing*, 26 (1), pp. 16–28.

Prochaska, J. O. and Velicer, W. F. (1997) The transtheoretical model of health behavior change, *American Journal of Health Promotion*, 12 (1), pp. 38–48.

Rasmussen, L. N. and Montgomery, P. (2018) The prevalence of and factors associated with inclusion of non-English language studies in Campbell systematic reviews: a survey and meta-epidemiological study, *Systematic Reviews*, 7 (1), pp. 129.

Razali, N. M. and Wah, Y. B. (2011) Power comparisons of shapiro-wilk, kolmogorov-smirnov, lilliefors and anderson-darling tests, *Journal of Statistical Modeling and Analytics*, 2 (1), pp. 21–33.

- Reed, F. M., Fitzgerald, L. and Bish, M. R. (2016) Mixing methodology, nursing theory and research design for a practice model of district nursing advocacy, *Nurse Researcher*, 23 (3), pp. 37–41.
- Rehm, R. S. (2013) Nursing's contribution to research about parenting children with complex chronic conditions: An integrative review, 2002 to 2012, *Nursing Outlook*, 61 (5), pp. 266–290.
- Rodgers, C., Bertini, V., Conway, M. A., Crosty, A., Filice, A., Herring, R. A., *et al.* (2018) A Standardized Education Checklist for Parents of Children Newly Diagnosed With Cancer: A Report From the Children's Oncology Group, *Journal of Pediatric Oncology Nursing*, pp. 1043454218764889.
- Roe, A. M. and Liberman, A. (2007) A comparative analysis of the United Kingdom and the United States health care systems, *The Health Care Manager*, 26 (3), pp. 190–212.  
DOI:10.1097/01.HCM.0000285010.03526.f5.
- Rogers, E. M. (1976) New product adoption and diffusion, *Journal of Consumer Research*, pp. 290–301.
- Rojas-Cooley, M. T. and Grant, M. (2009) Complementary and alternative medicine: oncology nurses' knowledge and attitudes, *Oncology Nursing Forum*, 36 (2), pp. 217–224.  
DOI:10.1188/09.ONF.217-224.
- Rolfe, G. (2006) Validity, trustworthiness and rigour: quality and the idea of qualitative research, *Journal of Advanced Nursing*, 53 (3), pp. 304–310. DOI:10.1111/j.1365-2648.2006.03727.x.
- Rossetto, K. R. (2014) Qualitative research interviews: Assessing the therapeutic value and challenges, *Journal of Social and Personal Relationships*, 31 (4), pp. 482–489.
- Rost, M., Wangmo, T., Niggli, F., Hartmann, K., Hengartner, H., Ansari, M., *et al.* (2017) Parents' and physicians' perceptions of children's participation in decision-making in paediatric oncology: A quantitative study, *Journal of Bioethical Inquiry*, 14 (4), pp. 555–565.
- Roth-Isigkeit, A., Thyen, U., Stöven, H., Schwarzenberger, J. and Schmucker, P. (2005) Pain among children and adolescents: restrictions in daily living and triggering factors, *Pediatrics*, 115 (2), pp. e152–e162.
- Royal College of Nursing (2009) Clinical Practice Guideline: The recognition and assessment of acute pain in children. London, England, RCN Institute. Available from: [http://www.rcpch.ac.uk/system/files/protected/page/RCN%20pain%20in%20children\\_publication\\_draft\\_30%2007%2009.pdf](http://www.rcpch.ac.uk/system/files/protected/page/RCN%20pain%20in%20children_publication_draft_30%2007%2009.pdf) [Accessed 3 January 2016].



- Roza, K. A., Horton, J. R., Johnson, K. and Anderson, W. G. (2014) Update in Hospice and Palliative Care, *Journal of Palliative Medicine*, 17 (3), pp. 351–357.
- Ruhe, K. M., Badarau, D. O., Brazzola, P., Hengartner, H., Elger, B. S., Wangmo, T. and Swiss Pediatric Oncology Group (2016) Participation in pediatric oncology: views of child and adolescent patients, *Psycho-Oncology*, 25 (9), pp. 1036–1042.
- Running, A. and Seright, T. (2012) Integrative oncology: managing cancer pain with complementary and alternative therapies, *Current Pain and Headache Reports*, 16 (4), pp. 325–331.
- Russo, M. W. (2007) How to Review a Meta-analysis, *Gastroenterology & Hepatology*, 3 (8), pp. 637–642.
- Sandelowski, M., Voils, C. I. and Knafl, G. (2009) On quantizing, *Journal of Mixed Methods Research*, 3 (3), pp. 208–222.
- Sato, I., Higuchi, A., Yanagisawa, T., Mukasa, A., Ida, K., Sawamura, Y., *et al.* (2014) Cancer-specific health-related quality of life in children with brain tumors, *Quality of Life Research*, 23 (4), pp. 1059–1068.
- Schmider, E., Ziegler, M., Danay, E., Beyrer, L. and Bühner, M. (2010) Is it really robust?, *Methodology*.
- Schonla, M. and Toepoel, V. (2015) Straightlining in Web survey panels over time, in: *Survey Research Methods*.9, pp. 125–137.
- Schreier, M. (2014) Qualitative content analysis, *The SAGE Handbook of Qualitative Data Analysis*, pp. 170–183.
- Schulz, K. F., Altman, D. G. and Moher, D. (2010) CONSORT 2010 statement: updated guidelines for reporting parallel group randomised trials, *BMC Medicine*, 8 (1), pp. 1.
- Schulz, K. F. and Grimes, D. A. (2005) Sample size calculations in randomised trials: mandatory and mystical, *The Lancet*, 365 (9467), pp. 1348–1353. DOI:10.1016/S0140-6736(05)61034-3.
- Senft, P. (1960) The dynamics of planned change, *International Journal of Social Psychiatry*, 5 (4), pp. 315–316.
- Sepponen, K., Kokki, H. and Ahonen, R. (1999) Training of medical staff positively influences postoperative pain management at home in children, *Pharmacy World & Science*, 21 (4), pp. 168–172.

- Shepherd, E., Woodgate, R. L. and Sawatzky, J.-A. (2010) Pain in children with central nervous system cancer: a review of the literature, *Oncology Nursing Forum*, 37 (4), pp. E318-330. DOI:10.1188/10.ONF.E318-E330.
- Silverman, D. (2009) *Doing Qualitative Research*. 3rd Revised edition. London; Los Angeles: SAGE Publications Ltd.
- Silverman, D. (ed.) (2010) *Qualitative Research*. 3rd Revised edition. Los Angeles: Sage Publications Ltd.
- Sinatra, R. (2010) Causes and Consequences of Inadequate Management of Acute Pain, *Pain Medicine*, 11 (12), pp. 1859–1871. DOI:10.1111/j.1526-4637.2010.00983.x.
- Singh, J. (2013) Critical appraisal skills programme, *Journal of Pharmacology and Pharmacotherapeutics*, 4 (1), pp. 76.
- Slevin, S. and Sines, S. (2000) Enhancing the truthfulness, consistency and transferability of a qualitative study: utilising a manifold of approaches, *Nurse Researcher*, 7 (2), pp. 79–98. DOI:10.7748/nr2000.01.7.2.79.c6113.
- Smith, J., Cheater, F. and Bekker, H. (2015) Parents' experiences of living with a child with a long-term condition: a rapid structured review of the literature, *Health Expectations*, 4 (18), pp. 452–474.
- Smith, J. and Noble, H. (2014) Bias in research, *Evidence-Based Nursing*, 17 (2), pp. 2–3.
- Söderbäck, M., Coyne, I. and Harder, M. (2011) The importance of including both a child perspective and the child's perspective within health care settings to provide truly child-centred care, *Journal of Child Health Care*, 15 (2), pp. 99–106.
- Stub, T., Quandt, S. A., Arcury, T. A., Sandberg, J. C. and Kristoffersen, A. E. (2018) Attitudes and knowledge about direct and indirect risks among conventional and complementary health care providers in cancer care, *BMC Complementary and Alternative Medicine*, 18 (1), pp. 44.
- Sutters, K. A., Holdridge-Zeuner, D., Waite, S., Paul, S. M., Savedra, M. C., Lanier, B., Mahoney, K. and Miaskowski, C. (2012) A descriptive feasibility study to evaluate scheduled oral analgesic dosing at home for the management of postoperative pain in preschool children following tonsillectomy, *Pain Medicine*, 13 (3), pp. 472–483. DOI:10.1111/j.1526-4637.2011.01324.x.
- Sutters, K. A., Miaskowski, C., Holdridge-Zeuner, D., Waite, S., Paul, S. M., Savedra, M. C. and Lanier, B. (2004) A randomized clinical trial of the effectiveness of a scheduled oral analgesic dosing regimen for the management of postoperative pain in children following tonsillectomy, *Pain*, 110 (1–2), pp. 49–55. DOI:10.1016/j.pain.2004.03.008.

- Sutters, K. A., Miaskowski, C., Holdridge-Zeuner, D., Waite, S., Paul, S. M., Savedra, M. C., Lanier, B. and Mahoney, K. (2010) A randomized clinical trial of the efficacy of scheduled dosing of acetaminophen and hydrocodone for the management of postoperative pain in children following tonsillectomy, *The Clinical Journal of Pain*, 26 (2), pp. 95–103.  
DOI:10.1097/AJP.0b013e3181b85f98.
- Svedberg, P., Einberg, E.-L., Wärnestål, P., Stigmar, J., Castor, A., Enskär, K. and Nygren, J. M. (2016) Support from healthcare services during transition to adulthood—Experiences of young adult survivors of pediatric cancer, *European Journal of Oncology Nursing*, 21, pp. 105–112.
- Teddlie, C. and Tashakkori, A. (2009) *Foundations of Mixed Methods Research: Integrating Quantitative and Qualitative Approaches in the Social and Behavioral Sciences*. SAGE Publications Inc.
- Teddlie, C. and Yu, F. (2007) Mixed methods sampling: A typology with examples, *Journal of Mixed Methods Research*, 1 (1), pp. 77–100.
- Tennant, F. (2013) The physiologic effects of pain on the endocrine system, *Pain and Therapy*, 2 (2), pp. 75–86.
- The Nemours Foundation (2018) Nutritional Needs for Kids With Cancer. Available from: <https://kidshealth.org/en/parents/cancer-nutrition.html> [Accessed 12 July 2018].
- Thomas, D. R. (2006) A general inductive approach for analyzing qualitative evaluation data, *American Journal of Evaluation*, 27 (2), pp. 237–246.
- Todd, A. (2010) Interviewing, in: Gerrish, K. and Lacey, A. (eds.) *The Research Process in Nursing*. John Wiley & Sons,.
- Tritter, J. Q. and McCallum, A. (2006) The snakes and ladders of user involvement: Moving beyond Arnstein, *Health Policy*, 76 (2), pp. 156–168. DOI:10.1016/j.healthpol.2005.05.008.
- Tutelman, P. R., Chambers, C. T., Stinson, J. N., Parker, J. A., Fernandez, C. V., Witteman, H. O., et al. (2018) Pain in children with cancer: prevalence, characteristics, and parent management, *The Clinical Journal of Pain*, 34 (3), pp. 198–206. DOI:10.1097/AJP.0000000000000531.
- Twycross, A. (1998) Children’s cognitive level and their perception of pain: children’s perception of pain changes as they develop understanding in the developmental stages, *Paediatric Nursing*, 10 (3), pp. 24–27. DOI:10.7748/paed.10.3.24.s21.
- Twycross, A. (2010) Managing pain in children: Where to from here?, *Journal of Clinical Nursing*, 19 (15–16), pp. 2090–2099.

Twycross, A., Forgeron, P. and Williams, A. (2015a) Paediatric nurses' postoperative pain management practices in hospital based non-critical care settings: A narrative review, *International Journal of Nursing Studies*, 52 (4), pp. 836–863. DOI:10.1016/j.ijnurstu.2015.01.009.

Twycross, A., Parker, R., Williams, A. and Gibson, F. (2015b) Cancer-related pain and pain management sources, prevalence and the experiences of children and parents, *Journal of Pediatric Oncology Nursing*, 32 (6), pp. 369–384.

Twycross, A., Voepel-Lewis, T., Vincent, C., Franck, L. and von Baeyer, C. (2015c) A debate on the proposition that self-report is the gold standard in assessment of pediatric pain intensity, *The Clinical Journal of Pain*, 31 (8), pp. 707–712. DOI:10.1097/AJP.000000000000165.

Twycross, A., Williams, A., Bolland, R. and Sunderland, R. (2015d) Parental attitudes to children's pain and analgesic drugs in the United Kingdom, *Journal of Child Health Care*, 19 (3), pp. 402–411. DOI:10.1177/1367493513517305.

Twycross, A., Williams, A. M., Bolland, R. E. and Sunderland, R. (2014) Parental attitudes to children's pain and analgesic drugs in the United Kingdom, *Journal of Child Health Care: For Professionals Working with Children in the Hospital and Community*. DOI:10.1177/1367493513517305.

Unger, J. B., Gallaher, P., Palmer, P. H., Baezconde-Garbanati, L., Trinidad, D. R., Cen, S. and Johnson, C. A. (2004) No news is bad news characteristics of adolescents who provide neither parental consent nor refusal for participation in school-based survey research, *Evaluation Review*, 28 (1), pp. 52–63. DOI:10.1177/0193841X03254421.

Unsworth, V., Franck, L. S. and Choonara, I. (2007) Parental assessment and management of children's postoperative pain: a randomized clinical trial, *Journal of Child Health Care*, 11 (3), pp. 186–194. DOI:10.1177/1367493507079558.

Van Cleve, L., Bossert, E., Beecroft P, Adlard K, Alvarez O and Savedra MC (2004) The pain experience of children with leukemia during the first year after diagnosis, *Nursing Research*, 53 (1), pp. 1- 10 10p.

Van Cleve, L., Muñoz, C. E., Savedra, M., Riggs, M., Bossert, E., Grant, M. and Adlard, K. (2012) Symptoms in children with advanced cancer: child and nurse reports, *Cancer Nursing*, 35 (2), pp. 115–125. DOI:10.1097/NCC.0b013e31821aedba.

Van Hout, M., Bergin, M., Foley, M., Rich, E., Rapca, A. I., Harris, R. and Norman, I. (2014) A scoping review of codeine use, misuse and dependence, *Final Report CODEMISUSED Project European Commission 7th Framework Programme, Brussels*. Available from:

[http://www.academia.edu/download/42060271/Van\\_Hout\\_MC\\_Bergin\\_M\\_Foley\\_M\\_Rich\\_E\\_20160204-27177-4ujzra.pdf](http://www.academia.edu/download/42060271/Van_Hout_MC_Bergin_M_Foley_M_Rich_E_20160204-27177-4ujzra.pdf) [Accessed 27 July 2017].

Versloot, J., Veerkamp, J. S. and Hoogstraten, J. (2004) Assessment of pain by the child, dentist, and independent observers, *Pediatric Dentistry*, 26 (5), pp. 445–449.

Versloot, J., von Baeyer, C. L. and Craig, K. D. (2013) Children give different self-reports of pain intensity to different people: the influence of social display rules, *Children*, 15 (2).

Vincent, C., Chiappetta, M., Beach, A., Kiolbasa, C., Latta, K., Maloney, R. and Van Roeyen, L. S. (2012) Parents' management of children's pain at home after surgery, *Journal for Specialists in Pediatric Nursing*, 17 (2), pp. 108–120. DOI:10.1111/j.1744-6155.2012.00326.x.

von Baeyer, C. L., Marche, T. A., Rocha, E. M. and Salmon, K. (2004) Children's memory for pain: overview and implications for practice, *The Journal of Pain*, 5 (5), pp. 241–249. DOI:10.1016/j.jpain.2004.05.001.

Walco, G. A., Conte, P. M., Labay, L. E., Engel, R. and Zeltzer, L. K. (2005) Procedural distress in children with cancer: self-report, behavioral observations, and physiological parameters, *The Clinical Journal of Pain*, 21 (6), pp. 484–490.

Wallander, J. L., Varni, J. W., Babani, L., Banis, H. T., DeHaan, C. B. and Wilcox, K. T. (1989) Disability parameters, chronic strain, and adaptation of physically handicapped children and their mothers, *Journal of Pediatric Psychology*, 14 (1), pp. 23–42.

Walsh, A., Edwards, H. and Fraser, J. (2009) Attitudes and subjective norms: determinants of parents' intentions to reduce childhood fever with medications, *Health Education Research*, 24 (3), pp. 531–545. DOI:10.1093/her/cyn055.

Walther-Larsen, S., Aagaard, G. B., Friis, S. M., Petersen, T., Møller-Sonnergaard, J. and Rømsing, J. (2016) Structured intervention for management of pain following day surgery in children, *Paediatric Anaesthesia*, 26 (2), pp. 151–157. DOI:10.1111/pan.12811.

Wang, J., Jacobs, S., Dewalt, D. A., Stern, E., Gross, H. and Hinds, P. S. (2017) A longitudinal study of PROMIS pediatric symptom clusters in children undergoing chemotherapy, *Journal of Pain & Symptom Management*, 54 (3), pp. 359–367. DOI:10.1016/j.jpainsymman.2017.08.021.

Ward, D. J., Furber, C., Tierney, S. and Swallow, V. (2013) Using framework analysis in nursing research: a worked example, *Journal of Advanced Nursing*, 69 (11), pp. 2423–2431.

Ward, E., DeSantis, C., Robbins, A., Kohler, B. and Jemal, A. (2014) Childhood and adolescent cancer statistics, 2014, *CA: A Cancer Journal for Clinicians*, 64 (2), pp. 83–103.

- Wengraf, T. (2001) *Qualitative research interviewing: Biographic narrative and semi-structured methods*. Sage.
- West, C. H., Bell, J. M., Woodgate, R. L. and Moules, N. J. (2015) Waiting to return to normal: An exploration of family systems intervention in childhood cancer, *Journal of Family Nursing*, 21 (2), pp. 261–294.
- Whittemore, R. and Knafl, K. (2005) The integrative review: updated methodology, *Journal of Advanced Nursing*, 52 (5), pp. 546–553. DOI:10.1111/j.1365-2648.2005.03621.x.
- WHO (2012) WHO | WHO guidelines on persisting pain in children, WHO. Available from: [http://www.who.int/medicines/areas/quality\\_safety/guide\\_perspainchild/en/](http://www.who.int/medicines/areas/quality_safety/guide_perspainchild/en/) [Accessed 28 January 2016].
- Wickham, R. (2007) Chemotherapy-induced peripheral neuropathy: a review and implications for oncology nursing practice, *Clinical Journal of Oncology Nursing*, 11 (3), pp. 361.
- Wiffen, P. J., Cooper, T. E., Anderson, A.-K., Gray, A. L., Grégoire, M.-C., Ljungman, G. and Zernikow, B. (2017) Opioids for cancer-related pain in children and adolescents, *The Cochrane Database of Systematic Reviews*, 7, pp. CD012564. DOI:10.1002/14651858.CD012564.pub2.
- Wiffen, P. J., Wee, B. and Moore, R. A. (2016) Oral morphine for cancer pain, *The Cochrane Database of Systematic Reviews*, 4, pp. CD003868. DOI:10.1002/14651858.CD003868.pub4.
- Wiggins, S. A. (2009) Family exemplars during implementation of a home pain management intervention, *Issues in Comprehensive Pediatric Nursing*, 32 (4), pp. 160–179. DOI:10.3109/01460860903281341.
- Williams, L. K. and McCarthy, M. C. (2015) Parent perceptions of managing child behavioural side-effects of cancer treatment: a qualitative study, *Child: Care, Health and Development*, 41 (4), pp. 611–619.
- Willig, C. (2013) *Introducing qualitative research in psychology*. 3rd ed. McGraw-Hill Education (UK).
- Wong, D. L. and Baker, C. M. (1988) Pain in children: comparison of assessment scales, *Pediatric Nursing*, 14 (1), pp. 9–17.
- Woodgate, R. L. and Degner, L. F. (2003) Expectations and beliefs about children's cancer symptoms: perspectives of children with cancer and their families, *Oncology Nursing Forum*, 30 (3), pp. 479–491. DOI:10.1188/03.ONF.479-491.

- Zalenski, R. J. and Raspa, R. (2006) Maslow's hierarchy of needs: a framework for achieving human potential in hospice, *Journal of Palliative Medicine*, 9 (5), pp. 1120–1127.
- Zaykin, D. V., Zhivotovsky, L. A., Westfall, P. H. and Weir, B. S. (2002) Truncated product method for combining P-values, *Genetic Epidemiology*, 22 (2), pp. 170–185.  
DOI:10.1002/gepi.0042.
- Zernikow, B., Hasan, C., Hechler, T., Huebner, B., Gordon, D. and Michel, E. (2008) Stop the pain! A nation-wide quality improvement programme in paediatric oncology pain control, *European Journal of Pain*, 12 (7), pp. 819–833. DOI:10.1016/j.ejpain.2007.12.002.
- Zernikow, B., Smale, H., Michel, E., Hasan, C., Jorch, N. and Andler, W. (2006) Paediatric cancer pain management using the WHO analgesic ladder—results of a prospective analysis from 2265 treatment days during a quality improvement study, *European Journal of Pain*, 10 (7), pp. 587–587.
- Zhou, H., Roberts, P. and Horgan, L. (2008) Association between self-report pain ratings of child and parent, child and nurse and parent and nurse dyads: meta-analysis, *Journal of Advanced Nursing*, 63 (4), pp. 334–342.
- Zhukovsky, D. S., Rozmus, C. L., Robert, R. S., Bruera, E., Wells, R. J., Chisholm, G. B., Allo, J. A. and Cohen, M. Z. (2015) Symptom profiles in children with advanced cancer: Patient, family caregiver, and oncologist ratings, *Cancer*, 121 (22), pp. 4080–4087. DOI:10.1002/cncr.29597.
- Zimmerman, B. J. (1986) Becoming a self-regulated learner: Which are the key subprocesses?, *Contemporary Educational Psychology*, 11 (4), pp. 307–313. DOI:10.1016/0361-476X(86)90027-5.
- Zisk, R. Y., Grey, M., MacLaren, J. E. and Kain, Z. N. (2007a) Exploring sociodemographic and personality characteristic predictors of parental pain perceptions, *Anesthesia & Analgesia*, 104 (4), pp. 790–798. DOI:10.1213/01.ane.0000257927.35206.c1.
- Zisk, R. Y., Grey, M., Medoff-Cooper, B. and Kain, Z. N. (2007b) Accuracy of parental-global-impression of children's acute pain, *Pain Management Nursing*, 8 (2), pp. 72–76.  
DOI:10.1016/j.pmn.2007.03.002.
- Zisk, R. Y. Z., Fortier, M. A., Chorney, J. M., Perret, D. and Kain, Z. N. (2010) Parental postoperative pain management: attitudes, assessment, and management, *Pediatrics*, 125 (6), pp. e1372-1378. DOI:10.1542/peds.2009-2632.

## **Appendices**



## Appendix 1: Characteristics of included studies

Author Date Country Design	Participants Condition	Aims	Intervention type, details and comparison	Statistical significance			Conclusions	Key points from risk of bias assessment
				Pain reductio n	Analgesic drug administration	Other outcomes		
Sutters et al. 2004 US RCT	Parents of children (n=80). Surgery: ENT	To determine whether ATC dosing, with or without nurse coaching, reduced children's reports of pain intensity, increased pain relief, and analgesic drug consumption.	ATC and nurse coaching 2 intervention groups both receiving digital timer: ATC group and ATC+Nurse coaching group Comparison: standard care	No	Yes (F (2, 77) = 24.55, P<0.001)	No significant difference in nausea and vomiting	Ineffective analgesic drugs meant that the intervention was ineffective. A surprising finding that the nurse coaching group was not significantly different. This is due to effective written instruction.	No demographic details provided of participants who withdrew.

Author Date Country Design	Participants Condition	Aims	Intervention type, details and comparison	Statistical significance			Conclusions	Key points from risk of bias assessment
				Pain reductio n	Analgesic drug administration	Other outcomes		
Wiggins et al. 2009 US RCT	Parents of children (n=13). Surgery: ENT	To describe how families implemented an alarm intervention designed to promote postoperative ATC administration of analgesic drugs.	ATC Asked to set an alarm as a reminder to administer prescribed analgesic drug. Comparison: standard care	No	Yes (by day 2) (range 4-6 doses for intervention group, 1-4 doses for control; p=0.014)	No significant difference in fluid intake or sleep	The intervention was ineffective due to heterogeneous and small sample, ineffective analgesic drugs.	Insufficient sample size. No provision of group homogeneity or demographic s provided.

Author Date Country Design	Participants Condition	Aims	Intervention type, details and comparison	Statistical significance			Conclusions	Key points from risk of bias assessment
				Pain reductio n	Analgesic drug administration	Other outcomes		
Sutters et al. 2010 US RCT	Parents of children (n=113). Surgery: ENT	To determine the effectiveness of ATC analgesic drug administration, with or without nurse coaching, compared to standard care.	ATC and nurse coaching 2 intervention groups both receiving digital timer: ATC group and ATC+Nurse coaching group Comparison: standard care	At rest: second evening t=2.23, p=0.028 ; second morning t=2.33, p=0.002 . With swallow ing: first morning t=2.11, p=0.037 ; second evening t=2.05, p=0.43; second	Yes (F(1,102)=49. 67, p<0.0001)	No significant difference in sedation, light- headedness, feeling dizzy, nightmares, nausea, vomiting, and constipation.	Effectiveness of intervention attributed to the combination of the specific analgesic drug chosen (acetaminophen with hydrocodone) and ATC dosing. Nurse coaching had no effect so written instructions are sufficient. ATC is appropriate because post-op pain is predictable. Pain intensity measured twice and fluctuations in	Insufficient power to detect small effect sizes. Significant differences between groups in mother's ethnicity.

Author Date Country Design	Participants Condition	Aims	Intervention type, details and comparison	Statistical significance			Conclusions	Key points from risk of bias assessment
				Pain reductio n	Analgesic drug administration	Other outcomes		
				morning t=2.99, p=0.003 ; third evening t=2.599, p=0.011			intensity not detected.	

Author Date Country Design	Participants Condition	Aims	Intervention type, details and comparison	Statistical significance			Conclusions	Key points from risk of bias assessment
				Pain reductio n	Analgesic drug administration	Other outcomes		
Chambers et al. 1997 Canada RCT	Parents (n=82). Surgery: ENT	To evaluate the effectiveness of a booklet for parents on the assessment and management of children's pain in terms of attitudes, assessment and medication administration.	Parent education Written information. Pain education booklet: "Pain, pain, go away: helping children with pain" Comparison: pain assessment control and no pain education control	No	Yes (only day 3) (mean = 0.8 [pain education group], 0.2 [assessment control group and no pain education group]; p<0.05)	Parents' attitudes (mean = 5.33 [pain education group], 4.82 [assessment control group], 4.76 no pain education group; p<0.01)	How parents assess pain and their attitudes toward children's pain medications contribute independently to how they medicate their children's pain. Optimal pain management achieved through targeting parents' concerns about pain medications and educating about assessment.	Randomization procedure is unclear. Slight variances in number and age of children of parents between groups. Small sample size may have prevented detection of small effects.

Author Date Country Design	Participants Condition	Aims	Intervention type, details and comparison	Statistical significance			Conclusions	Key points from risk of bias assessment
				Pain reductio n	Analgesic drug administration	Other outcomes		
Allen & Shriver 1998 US RCT	Children (n=27) and their parents. Migraine	To evaluate the efficacy of parent-mediated pain behaviour management strategies implemented by parents of children undergoing biofeedback treatment for migraine headache.	Parent education Biofeedback combined with written and verbal information on influence of parents' behaviour on child pain. Comparison: biofeedback alone.	Yes at 3 months (mean differen ce 1.4; p≤0.05) but not at 1 year	n/a not assessed	Adaptive functioning (mean difference 16, p≤0.05)	Success cannot be attributed to the intervention because there was no measure of adherence to the intervention. Significance only noted over first 3 months. Potentially participants forgot the intervention after that time. As both groups improved, the intervention may have meant participants reached the stage of recovery quicker than the control.	No indication randomizatio n procedure Small sample size may have prevented small effects to be detected at later time points. Therapist not blinded.

Author Date Country Design	Participants Condition	Aims	Intervention type, details and comparison	Statistical significance			Conclusions	Key points from risk of bias assessment
				Pain reductio n	Analgesic drug administration	Other outcomes		
Vincent et al. 2012 US Quasi- experim ental	Children (n=108) and their parents. Surgery: various	To compare the effectiveness of Home Pain Management for Children with usual discharge teaching on children's pain intensity, parents' analgesic drug administration, parents' and children's satisfaction with pain levels, and use of unplanned healthcare.	Parent education Written and verbal information. Pain Management Information sheet and follow up session to discuss sheet. Comparison: standard care	No	No	Child satisfaction ( $\chi^2=4.90$ , p=0.03)	The intervention was ineffective potentially due to: too much written information, insufficient emphasis on the importance of analgesic drugs, parents' attitudinal barriers, or small sample sizes.	Insufficient sample size. Sample mostly white children of educated parents.

Author Date Country Design	Participants Condition	Aims	Intervention type, details and comparison	Statistical significance			Conclusions	Key points from risk of bias assessment
				Pain reductio n	Analgesic drug administration	Other outcomes		
Helgado ttr & Wilson 2014 Iceland RCT	Children (n=93) and their parents. Surgery: ENT	To determine the effectiveness of educating parents to provide distraction in decreasing postoperative pain at home.	Parent education Written and verbal pain management and distraction education. Comparison: written pain management education only.	No	n/a not assessed	Pain behaviour (p=0.023; ηp <sup>2</sup> =.076)	Pain behaviour is an equally important aspect of pain experience as pain intensity. It was measured over a day as opposed to at one moment. Many children had clinically significant pain despite correct analgesic drug administration.	Block randomizatio n is not true randomizatio n.
Bailey et al. 2015 Australi a RCT	Parents of children (n=58). Surgery: ENT	To evaluate paediatric post- tonsillectomy pain management using	Parent education Written information. Oxycodone information	Yes, on day 3 (mean differen ce=1.07, p=0.05)	n/a not assessed	Parents' satisfaction (mean difference 1.69, p<0.001),	Correlational only, cannot say info sheet caused improvement. Having more effective analgesic drugs administered	Uneven gender distribution between groups.



Author Date Country Design	Participants Condition	Aims	Intervention type, details and comparison	Statistical significance			Conclusions	Key points from risk of bias assessment
				Pain reductio n	Analgesic drug administration	Other outcomes		
		oxycodone when a specific analgesic drug information sheet is provided.	sheet Comparison: standard care	and day 7 (mean differen ce=1.55, p=0.02), not on day 5		parents' knowledge ( $\chi^2=29.53$ , p<0.001)	and better analgesic drug control.	

Author Date Country Design	Participants Condition	Aims	Intervention type, details and comparison	Statistical significance			Conclusions	Key points from risk of bias assessment
				Pain reductio n	Analgesic drug administration	Other outcomes		
Unsworth et al. 2007 UK RCT	Parents of children (n=88). Surgery: ENT	To determine whether the use of a self-report pain scale would result in children receiving more analgesic drugs.	Provision of pain assessment tool Wong-Baker scale to determine child pain intensity Comparison: standard care	No	No	Improved administration of codeine as instructed (24% codeine administration [control], 37% [intervention]; p=0.004) and reduced inappropriate administration of analgesic drugs (69% control v 39% in intervention; p=0.001).	Intervention is ineffective. This may have been due to small sample size.	Insufficient sample size.

Author Date Country Design	Participants Condition	Aims	Intervention type, details and comparison	Statistical significance			Conclusions	Key points from risk of bias assessment
				Pain reductio n	Analgesic drug administration	Other outcomes		
Franck et al. 2007 UK RCT	Parents of children (n=25). Surgery: various	To determine whether parents' pain assessment documentation and analgesic drug administration increased with the use of a temporary tattoo of a pain intensity scale.	Provision of pain assessment tool Temporary tattoo Children given temporary tattoo of pain assessment scale Comparison: fun tattoo and paper pain scale.	No	No	More pain assessments at day 1 (3.0±1.16 vs. 1.93±.88; P<0.05).	Intervention may be helpful but alone is insufficient. Conclusions limited by lack of power.	Pilot study, underpowere d and not designed to consider the unanticipated effect of PCA / EA and or PCS. Heterogeneo us samples from different wards. Block randomizatio n.

Author Date Country Design	Participants Condition	Aims	Intervention type, details and comparison	Statistical significance			Conclusions	Key points from risk of bias assessment
				Pain reductio n	Analgesic drug administration	Other outcomes		
Kankku nen et al. 2009 Finland RCT	Parents (n=50). Surgery: unspecified	To evaluate the influence of parents' use of Parents' Postoperative Pain Measure on the use of pain medication at home.	Provision of pain assessment tool. Parents Postoperative Pain Management tool provided to parents. Comparison: standard care.	No	No	No differences in problems faced by parents	Parents who had pain assessment tool may have been more aware of pain but did not act on it. This intervention alone is insufficient.	Alternated allocation isn't randomizatio n. Potentially underpowere d and small sample size led to non- significant findings. No demographic details provided of participants who withdrew.

Author Date Country Design	Participants Condition	Aims	Intervention type, details and comparison	Statistical significance			Conclusions	Key points from risk of bias assessment
				Pain reductio n	Analgesic drug administration	Other outcomes		
Sutters et al. 2012 US Mixed methods	Parents of children (n=47). Surgery: ENT	To evaluate the feasibility of scheduled analgesic drug dosing following outpatient tonsillectomy to optimize pain management.	Multifaceted intervention Education on assessment, ATC instruction, provision of timer, written information, follow up phone calls, nurse coaching. Comparison: no control	Yes but no control	Yes but no control	Side-effects, sleep and oral intake measured but no control group	Effectiveness of intervention attributed to the combination of the specific analgesic drug chosen and ATC dosing. Daytime sedation, nausea and constipation were side-effects. Sedative effects of analgesic drug may have led to lower pain scores. Pain intensity only measured twice. Results may be due to surgical technique.	No control group prevents any meaningful conclusions of intervention efficacy.

Author Date Country Design	Participants Condition	Aims	Intervention type, details and comparison	Statistical significance			Conclusions	Key points from risk of bias assessment
				Pain reductio n	Analgesic drug administration	Other outcomes		
Walther -Larsen et al. 2016 Denmar k Prospect ive observat ional cohort	Parents of children (n=149). Surgery: various	To determine postoperative pain intensity after a structured intervention for pain management.	Multifaceted intervention Pain assessment tool, tailored provision of analgesic drugs, and parents' education written and verbal. Comparison: no control	Yes but no control	No	n/a	The intervention was successful, but the authors are not able to hone in on a specific aspect leading to the success.	No control group prevents any meaningful conclusions of intervention efficacy.

Author Date Country Design	Participants Condition	Aims	Intervention type, details and comparison	Statistical significance			Conclusions	Key points from risk of bias assessment
				Pain reductio n	Analgesic drug administration	Other outcomes		
Sepponen et al. 1999 Finland Pre and post experim ental	Parents (n=227). Surgery: ENT	To describe how parents manage their child's postoperative pain at home following day- case surgery.	Doctor and nurse education Staff training programme to improve analgesic drug medication practices, 1 hr lecture, 2 weeks bedside teaching. Comparison: pre- intervention parents	No	Yes, analgesic drug administration increased from 68% pre- intervention to 80% post intervention (p=0.028).	Ibuprofen use increased from 28% pre- intervention to 52% post intervention (p=0.002), acetaminophen sig decreased from 56% pre- intervention to 24% post intervention (p<0.001).	The training hospital staff improved written and verbal information supplied to parents. Use of suppositories is discussed.	Inferential statistics are not described. Methodology may have caused the study to be confounded by recall and social desirability bias.

Author Date Country Design	Participants Condition	Aims	Intervention type, details and comparison	Statistical significance			Conclusions	Key points from risk of bias assessment
				Pain reductio n	Analgesic drug administration	Other outcomes		
Palermo et al. 2009 US RCT	Children (n=48) and their parents. Chronic pain	To evaluate a more accessible treatment approach for chronic paediatric pain using an Internet- delivered family CBT intervention.	Family CBT via internet Web-based Management of Adolescent Pain: 2 websites (child and parent). 3 sections: homepage, treatment modules, daily diary. Comparison: wait list control.	Yes (mean differen ce 1.17, p=0.03)	n/a not assessed	Activity limitations (mean difference 2.74, p=0.004). No significant difference on depressive symptoms, parents' response, treatment acceptability, satisfaction.	Therapy was successful due to relatively high dose of parents' involvement and provision of parent strategies (as opposed to primarily child involvement).	Block randomizatio n is not true randomizatio n although blocks were allocated using random number generator.



Author Date Country Design	Participants Condition	Aims	Intervention type, details and comparison	Statistical significance			Conclusions	Key points from risk of bias assessment
				Pain reductio n	Analgesic drug administration	Other outcomes		
Hegarty et al. 2013 Australia RCT	Children (n=181) and their parents. Surgery: various	To investigate whether issuing parents with take-home analgesic drugs would improve postoperative pain scores and/or parents' satisfaction.	Hospital supplied analgesic drugs Parents supplied with take home hospital supplied analgesic drugs. Comparison: parent supplied analgesic drugs.	No	No	No differences in nausea, vomiting or sleep	Parents already have medications at home so providing them does not make a difference to effective pain management. Other barriers to effective pain management exist and should be investigated.	Block randomizatio n is not true randomizatio n. Homogeneity of groups not assessed, and demographic s not provided. Reasons for withdrawals not provided.

Author Date Country Design	Participants Condition	Aims	Intervention type, details and comparison	Statistical significance			Conclusions	Key points from risk of bias assessment
				Pain reductio n	Analgesic drug administration	Other outcomes		
Paquette et al. 2013 Canada RCT	Parents of children (n=45). Surgery: ENT	To determine if a nurse telephone follow-up with parents could decrease pain intensity, incidence of postoperative complications, and additional healthcare resource use.	Nurse telephone follow up Phone call on days 1,3,5 and 10 to provide support and information. Comparison: standard care	No	Yes, at day 1 ( $\chi^2(1) = 6.429$ , P=0.01) and day 3 ( $\chi^2(1) =$ 9.911, P=0.002)	Increased constipation at day 3 ( $\chi^2(1) =$ 13.672, P<0.001) and fluid intake at day 1( $\chi^2(1) =$ 7.202, P=0.007) and 3 ( $\chi^2(1) =$ 5.909, P=0.015). No significant difference in vomiting, fever, dizziness.	The intervention was ineffective. Potentially due to insufficient analgesic drug power, insufficient sample size, or difference in samples pre- intervention.	Underpowered. Significant group difference in vomiting pre- intervention so groups may not have been equal to start.

## **Appendix 2: Participant information sheet - survey**



### **An exploration of how parents manage their child's cancer-related pain at home**

Thank you for taking the time to read this information sheet. This information sheet is 4 pages long, please ensure you have all of the pages.

You are being invited to take part in a research study. Before you decide whether to participate, it is important for you to understand why the research is being done and what it will involve.

Please take time to read the following information carefully. Feel free to talk to others about the study if you wish. Please feel free to ask if there is anything that is not clear or if you would like more information.

Participation will not affect the care of your child. Participation is voluntary, and you are free to withdraw at any point in the study and you do not need to give a reason. Take time to decide whether or not you wish to participate.

You will then be given a copy of the survey to complete at your convenience when you are ready. If you do not wish to complete it, you do not need to do anything further.

### **What is this research about?**

- Children with cancer can experience pain throughout their cancer journey.
- Children with cancer now have more of their cancer care at home and less in hospital. This increases quality of life but means that it is important to help parents with the challenges of managing pain away from the hospital.
- Parents have told us that they find this challenging and want more help.
- This area is under-studied worldwide and has never been studied in the UK.
- There's a lot about the cancer journey that we can't fix, pain is something that we can fix.
- The overall aim of this research is to explore how parents of children with cancer manage their child's cancer pain at home with the goal of developing an intervention to support parents in managing their child's cancer pain.

### **What does this study involve?**

- This study has 3 phases:
  - o Survey

- Pain diary
- Interview
- Which phases will I be involved with?
  - You can choose to participate in as many or as few of the phases as you want. (You don't have to participate in the study at all.)

## **Survey**

- The survey asks you about pain in children and medications which can be given to children when they are in pain.
- The survey will take approximately 15 minutes to complete.

## **Pain diary**

- The pain diary asks you to keep a record for one month of your child's pain and your actions in response to their pain.
- Each day you will be asked to record their pain in the morning and the evening. Each recording will take approximately 5 minutes.

## **Interview**

- The interview will discuss barriers and facilitators to pain management at home.
- It will last approximately 1 hour.

## **Where will the research take place?**

The recruitment will take place in McElwain ward and Children's Day Unit at the Royal Marsden Hospital in Sutton. You can choose where you would like to complete the survey and the interview. The pain diary will take place whilst you and your child are at home.

## **When will the research take place?**

The research is being conducted from Autumn 2016 until Spring 2018. You will be able to choose when you complete each phase of the research.

## **What if something goes wrong?**

In this type of study, it is unlikely that anything will go wrong, however if you have a concern about any aspect of this study you should speak to Theresa Wiseman ([Theresa.Wiseman@rmh.nhs.uk](mailto:Theresa.Wiseman@rmh.nhs.uk)). If you remain unhappy and wish to complain formally you can do this through the NHS Complaints Procedure. Details can be obtained from the Patient Advisory Liaison Service (PALS) in the hospital.

## **What are the possible risks and benefits?**

We do not expect there to be any significant risks to you involved in your participation. The cost to you will be your time.

There are no immediate benefits for you taking part in this study. The findings may contribute to a greater understanding of how to support parents managing their child's cancer related pain at home in the future. It is hoped that what is learned will be of benefit to other children with cancer in the future.

## **What will happen if I don't want to carry on with the study?**

You may withdraw from the study at any point and without giving a reason. The care of your child will not be affected whether you participate or not. If you wish to withdraw, please inform the researcher, Roses Parker ([Parker11@lsbu.ac.uk](mailto:Parker11@lsbu.ac.uk)).

## **Will my information be kept confidential?**

We will not collect personal information during this phase, so this phase is anonymous. If you are going to participate in future phases, we will need to be able to link your responses in which case, we will collect identifiable information which will be kept confidential and only the research team will see this information. Any publications will collate this information, so you will not be identifiable to yourself or others.

The information will be stored in a computer, which is password-protected. All identifiable information will be removed from the data and your record will be given a unique identifier. Only one file will link you to your unique identifier and this file will be password protected. Only the researcher will have access to this file and only the research team will have access to the whole dataset.

## **What will happen to the results of this study?**

This research will be published in nursing and medical journals and presented at conferences.

Please note that your responses will be reviewed and analysed as part of the study only. If you have any concerns about your child's pain, please contact your clinical team.

## **Who are the researchers?**

Roses Parker is a full-time PhD student who also works part-time as a children's cancer nurse. She will be providing you with what you need to participate and will be conducting the interviews. She

is supervised by Professor Alison Twycross, Professor Theresa Wiseman and Dr Stephen McKeever.

Professor Theresa Wiseman is the Principal Investigator who will take responsibility for this study. She is a Clinical Professor of Applied Health Research in Cancer Care at the university of Southampton and The Royal Marsden NHS Foundation Trust. Theresa holds a clinical academic appointment, which means she combines clinical practice and research.

## **Who is organising and funding this study?**

This study is organised by PhD student and children's nurse at the Royal Marsden Hospital Roses Parker. She is funded by a scholarship from London South Bank University and is supervised both clinically and academically. The research is sponsored by Royal Marsden NHS Foundation Trust. Insurance will be provided by the NHS indemnity scheme.

## **How have patients and the public been involved in this study?**

Parents who have experience caring for a child with cancer have been involved in designing this study by making sure all the documents are understandable and relevant.

## **Who has reviewed this study?**

This study has been reviewed and approved by the Royal Marsden's Committee for Clinical Research (CCR), and the Health Research Authority (HRA) North of Scotland (1) Research Ethics Committee (16/NS/01).

## **Further information and contact details**

For further information, please contact the Principle Investigator for this study: Theresa Wiseman, 020 7811 8516, Theresa.Wiseman@rmh.nhs.uk.

## An exploration of how parents manage their child's cancer-related pain at home

Thank you for participating in this research. This research will help parents in the future to manage their child's cancer-related pain at home.

Your time and input is gratefully received.

Attached is a survey which should take approximately 15 minutes to complete.

Your response will be kept anonymous and confidential.

The first section of this survey asks you to give your opinion on expression of pain in children and using pain medications in children.

Please circle the number you feel best corresponds with your belief.

		Strongly disagree	Disagree	Slightly disagree	Not sure	Slightly agree	Agree	Strongly agree
1	Children always express pain by crying or whining	1	2	3	4	5	6	7
2	Children always tell their parents when they are in pain	1	2	3	4	5	6	7
3	Children who are quiet are not in pain	1	2	3	4	5	6	7
4	Children who are playing are not in pain	1	2	3	4	5	6	7
5	Children experiencing pain report it immediately	1	2	3	4	5	6	7
6	Children exaggerate pain	1	2	3	4	5	6	7
7	Children complain about pain to get attention	1	2	3	4	5	6	7
8	Children feel less pain than adults	1	2	3	4	5	6	7
9	Children in pain have trouble sleeping	1	2	3	4	5	6	7
10	Children should be given pain medication as little as possible because of side effects	1	2	3	4	5	6	7

		Strongly disagree	Disagree	Slightly disagree	Not sure	Slightly agree	Agree	Strongly agree
11	Children who take pain medication for pain may learn to take drugs to solve other problems	1	2	3	4	5	6	7
12	Pain medication works the same no matter how often it is used	1	2	3	4	5	6	7
13	Pain medication works best when it is given as little as possible	1	2	3	4	5	6	7
14	Pain medication has many side effects	1	2	3	4	5	6	7
15	Children will become addicted to pain medication if they take it for pain	1	2	3	4	5	6	7
16	There is little need to worry about side-effects from pain medication	1	2	3	4	5	6	7
17	It is unlikely a child will become addicted to pain medication if taken for pain	1	2	3	4	5	6	7
18	Pain medication is addictive	1	2	3	4	5	6	7
19	Pain medication works best if saved for when the pain is quite bad	1	2	3	4	5	6	7
20	Using pain medication for children's pain leads to later drug abuse	1	2	3	4	5	6	7
21	There is little risk of addiction when pain medication is given for pain	1	2	3	4	5	6	7
22	Children learn how to use pain medication responsibly when it is given for pain	1	2	3	4	5	6	7
23	Side effects are something to worry about when giving children pain medication	1	2	3	4	5	6	7
24	The less often children take pain medication for pain, the better the medicine works	1	2	3	4	5	6	7
25	Giving children pain medication for pain teaches proper use of drugs	1	2	3	4	5	6	7



The second section of this survey asks some demographic information which will help contextualise your responses.

### Questions about your child

How old is your child?	
What is your child's gender?	Male / Female (please circle)
Please circle one option which best describes your child's ethnic group or background:	<ul style="list-style-type: none"> <li>• White</li> <li>• Mixed / Multiple ethnic groups</li> <li>• Asian / Asian British</li> <li>• Black / African / Caribbean / Black British</li> <li>• Other ethnic group (please describe) _____</li> </ul>
What is your child's diagnosis?	
How long ago was your child diagnosed?	
What is your child's local hospital for cancer care?	

### Questions about you

What is your relationship to your child?	Mother / Father / Other (please describe) _____
What is your age? (please circle)	<input type="checkbox"/> 18-24 years <input type="checkbox"/> 25-34 years <input type="checkbox"/> 35-44 years <input type="checkbox"/> 45-54 years <input type="checkbox"/> 55-64 years <input type="checkbox"/> 65 years or older
Please circle one option which best describes your ethnic group or background:	<input type="checkbox"/> White <input type="checkbox"/> Mixed / Multiple ethnic groups <input type="checkbox"/> Asian / Asian British <input type="checkbox"/> Black / African / Caribbean / Black British <input type="checkbox"/> Other ethnic group (please describe) _____
What is your annual household income? (please circle)	<input type="checkbox"/> Less than £14,000 per year <input type="checkbox"/> £15,000 – £24,000 per year <input type="checkbox"/> £25,000 – £39,000 per year <input type="checkbox"/> £40,000 – £59,000 per year <input type="checkbox"/> More than £60,000 per year
Please circle on which best describes your educational level:	<input type="checkbox"/> Didn't finish school <input type="checkbox"/> Finished school <input type="checkbox"/> Certificate or partial studies at college/universities <input type="checkbox"/> Completed a Bachelor's degree <input type="checkbox"/> Completed a postgraduate degree
Do you have health-related education? (please circle)	Yes / No
If yes, please describe:	

Thank you!

You have reached the end of the survey. Thank you for your time, this research could not happen without you. Your responses will help parents to manage their child's cancer pain in the future. If you have any questions or points for clarification, please feel free to contact me: [parker11@lsbu.ac.uk](mailto:parker11@lsbu.ac.uk).

Instructions for return

Please return this form via the collection boxes in ward areas. Your nurse today should know where they are. Alternatively, please return via post using the stamped, addressed envelope which was provided to you with this survey.

Once again, many thanks for your help.

Would you like to receive a copy of the results when the research is finished?

If so, please send an email to [parker11@lsbu.ac.uk](mailto:parker11@lsbu.ac.uk) stating this. The research is likely to be finished towards the end of 2018.

Would you like to participate in the next phase?

If you would like to participate in another phase of the research please complete this form and return it either to the collection boxes in ward areas or via post using the stamped, addressed envelope which was provided to you with this survey. The researcher will be in contact with you shortly after the receipt of this form. This research is entirely voluntary, and you may withdraw at any point.

Name			
Email address			
Phone number		Mobile number	
Preferred mode of contact		Preferred time of contact	
I am interested in (please tick)	<input type="checkbox"/> Pain diary	<input type="checkbox"/> Interview	<input type="checkbox"/> Both

## **Appendix 4: Participant information sheet – pain diary and interview**



### **An exploration of how parents manage their child's cancer-related pain at home**

Thank you for taking the time to read this information sheet. This information sheet is 4 pages long, please ensure you have read all the pages.

You are being invited to take part in the next stage of a research study because you have already completed the first stage – a survey.

Before you decide whether to participate further, it is important for you to understand why the research is being done and what it will involve.

You are free to withdraw at any point in the study and you don't need to give a reason. Participation will not affect the care of your child.

Feel free to talk to others about the study if you wish. Please feel free to ask if there is anything that is not clear or if you would like more information. Take time to decide whether or not you wish to participate.

### **What is this research about?**

- Children with cancer can experience pain throughout their cancer journey.
- Children with cancer are now spending more of their cancer care at home and less in hospital. This increases quality of life but means that it is important to help parents with the challenges of managing pain away from the hospital.
- Parents have told us that they find this challenging and want more help.
- This area is under-studied worldwide and has never been studied in the UK.
- There's a lot about the cancer journey that we can't fix, pain is something that we can fix.
- The overall aim of this research is to explore how parents of children with cancer manage their child's cancer pain at home with the goal of developing an intervention to support parents in managing their child's cancer pain.

### **What does this study involve?**

This study has 3 phases:

- Survey
- Pain diary

- Interview

You have already had opportunity to participate in the survey. You can choose whether to continue your participation by completing a pain diary and / or an interview

## **Tell me about the pain diary**

### **What format will the pain diary be in?**

You will be given either a paper booklet or an online link to a pain diary. You can choose which one.

### **What do I have to do?**

Every morning and evening over a one-month period you will be asked to record:

- Presence of pain
- Intensity of pain
- Location of pain
- What you think might have caused the pain
- Your actions in response to pain
- The outcome of your actions

The diary also includes a space for comments. If your child experiences a pain episode in between the morning and evening recording times, you can record it in this space.

### **What are the possible benefits and risks?**

There are no direct benefits to you or your child. We are unable to offer any payment for your participation. However, the information you provide may help us improve the support we give to parents in the future.

## **Tell me about the interview**

If you choose to participate in the interview, a researcher will meet with you face-to-face or talk over the phone if you prefer. The interview will take place either in your home, the researchers' office or in a public place such as a cafe, whichever is most convenient. The interview will discuss what helps and what doesn't help when managing your child's pain. The interview will last approximately 60 minutes and will be audio-recorded. During this time, you can ask to stop or pause or a break if you feel you need it.

### **What are the possible benefits and risks?**

There are no direct benefits to you or your child. We are unable to offer any payment for your participation. However, the information you provide may help us improve the support we give to parents in the future.

Some people find research interviews like this one to be helpful in a therapeutic way. Some people find these types of interviews upsetting. It may be that we cover topics which are difficult to talk about. If you do become upset, there are support services available via your child's healthcare team, psychology services and charities like CLIC Sargent.

## **Where will the research take place?**

The research is based in McElwain ward and Children's Day Unit at the Royal Marsden NHS Foundation Trust in Sutton. You can choose where you would like to complete the interview. The pain diary will take place during a period of time you and your child are at home.

## **When will the research happen?**

The research is being conducted from Autumn 2016 until Spring 2018. You will be able to choose when you complete the interview. The pain diary will need to be completed during a period of time when your child is at home.

## **What if something goes wrong?**

In this type of study, it is unlikely that anything will go wrong, however if you have a concern about any aspect of this study you should speak to Theresa Wiseman ([Theresa.Wiseman@rmh.nhs.uk](mailto:Theresa.Wiseman@rmh.nhs.uk)). If you remain unhappy and wish to complain formally you can do this through the NHS Complaints Procedure. Details can be obtained from the Patient Advisory Liaison Service (PALS) in the hospital.

## **What will happen if I don't want to carry on with the study?**

You may withdraw from the study at any point and without giving a reason. The care of your child will not be affected whether you participate or not. If you wish to withdraw, please inform the researcher (Roses Parker: [Parker11@lsbu.ac.uk](mailto:Parker11@lsbu.ac.uk)).

## **Will my information be kept confidential?**

Pain diaries: The information collected in this phase will enable the researcher to identify you. This information will be kept confidential and only the research team will see this information. Any publications will collate this information, so you will not be identifiable to yourself or others. If your pain diaries reveal to me something which makes me think that you, or someone else's safety

is at risk, I may have to break your confidentiality. I would normally discuss this with you before doing this.

Interview: The information collected in this phase will enable the researcher to identify you. This information will be kept confidential and only the research team will see this information. Any publications will remove identifiable information so whilst you may be able to identify yourself in quotations used, no-one else will be able to identify you. If you say something to me which makes me think that you, or someone else's safety is at risk, I may have to break your confidentiality. I would normally discuss this with you before doing this.

The information will be stored in a computer, which is password-protected. All identifiable information will be removed from the data and your record will be given a unique identifier. Only one file will link you to your unique identifier and this file will be password protected. Only the researcher will have access to this file and only the research team will have access to the whole dataset.

Please note that your responses will be reviewed and analysed as part of the study only. If you have any concerns about your child's pain, please contact your clinical team.

## **What will happen to the results of this study?**

This research will be published in nursing and medical journals and presented at conferences.

## **Who are the researchers?**

Roses Parker is a full-time PhD student who also works part-time as a children's cancer nurse. She will be providing you with what you need to participate and will be conducting the interviews. She is supervised by Professor Alison Twycross, Professor Theresa Wiseman and Dr Stephen McKeever.

Professor Theresa Wiseman is the Principal Investigator who will take responsibility for this study. She is a Clinical Professor of Applied Health Research in Cancer Care at the university of Southampton and The Royal Marsden NHS Foundation Trust. Theresa holds a clinical academic appointment, which means she combines clinical practice and research.

## **Who is organising and funding this study?**

This study is organised by PhD student and children's nurse at the Royal Marsden Hospital, Roses Parker. She is funded by a scholarship from London South Bank University and is supervised both clinically and academically. The research is sponsored by Royal Marsden NHS Foundation Trust. Insurance will be provided by the NHS indemnity scheme.

## **How have patients and the public been involved in this study?**

Parents who have experience caring for a child with cancer have been involved in designing this study by making sure all the documents are understandable and relevant.

## **Who has reviewed this study?**

This study has been reviewed and approved by the Royal Marsden's Committee for Clinical Research (CCR), and the Health Research Authority (HRA) North of Scotland (1) Research Ethics Committee (16/NS/01).

## **Further information and contact details**

For further information, please contact the Principle Investigator for this study: Theresa Wiseman, 020 7811 8516, [Theresa.Wiseman@rmh.nhs.uk](mailto:Theresa.Wiseman@rmh.nhs.uk).





# An exploration of how parents manage their child's cancer-related pain at home

*The* ROYAL MARSDEN  
NHS Foundation Trust

## Consent form – Pain diary – Researcher copy

To indicate consent, please initial in the box

I have read and been given a copy of the participant information sheet (Version 1, 26/09/16) which describes the research in which I have been asked to participate.

The researcher has explained the nature and purpose of the research and I believe that I understand what is being proposed.

I have been given the opportunity to ask questions about the study and have received answers to my satisfaction.

I understand that taking part in the study will not affect the care of my child.

I understand that taking part is voluntary and that I may withdraw at any time and do not have to provide a reason for withdrawing.

I have been informed about what the data collected will be used for, to whom it may be disclosed, and how long it will be retained.

I hereby consent to participate in the study that has been fully explained to me.


-----

Name	Date	Signature
------	------	-----------

As the Researcher responsible for this study I confirm that I have explained to the participant named above the nature and purpose of the research to be undertaken.

-----

Researcher	Date	Signature
------------	------	-----------

If you are at all concerned about this study, please contact: Roslyn Parker, [parker11@lsbu.ac.uk](mailto:parker11@lsbu.ac.uk)

### Questions about you

What is your relationship to your child?	Mother / Father / Other (please describe)
What is your age? (please circle)	<input type="checkbox"/> 18-24 years <input type="checkbox"/> 25-34 years <input type="checkbox"/> 35-44 years <input type="checkbox"/> 45-54 years <input type="checkbox"/> 55-64 years <input type="checkbox"/> 65 years or older
Please tick one option which best describes your ethnic group or background:	<input type="checkbox"/> White <input type="checkbox"/> Mixed / Multiple ethnic groups <input type="checkbox"/> Asian / Asian British <input type="checkbox"/> Black / African / Caribbean / Black British <input type="checkbox"/> Other ethnic group (please describe) _____
What is your annual household income? (please tick)	<input type="checkbox"/> Less than £14,000 per year <input type="checkbox"/> £15,000 – £24,000 per year <input type="checkbox"/> £25,000 – £39,000 per year <input type="checkbox"/> £40,000 – £59,000 per year <input type="checkbox"/> More than £60,000 per year
Please tick which best describes your educational level:	<input type="checkbox"/> Didn't finish school <input type="checkbox"/> Finished school <input type="checkbox"/> Certificate or partial studies at college/universities <input type="checkbox"/> Completed a bachelor's degree <input type="checkbox"/> Completed a postgraduate degree
Do you have health-related education? (please circle)	Yes / No
If yes, please describe:	

### Questions about your child

How old is your child?	
What is your child's gender?	Male / Female (please circle)
Please circle one option which best describes your child's ethnic group or background:	<ul style="list-style-type: none"> <li>• White</li> <li>• Mixed / Multiple ethnic groups</li> <li>• Asian / Asian British</li> <li>• Black / African / Caribbean / Black British</li> <li>• Other ethnic group (please describe) _____</li> </ul>
What is your child's diagnosis?	
How long ago was your child diagnosed?	
Which hospitals are involved in your child's cancer care?	

## Appendix 6: Pain diary

1. Date:	Morning time:	Evening time:	Other episodes, time:
<b>Pain score out of 10</b> (0 = no pain; 10 = worst pain)			
<b>Location of pain</b>			
<b>What do you think caused the pain?</b>			
<b>What did you do in response to the pain?</b>			
<b>What happened to the pain once you tried to reduce it?</b>			
<b>If medicines were given:</b>			
<b>Name of medicines given</b>			
<b>Dose of medication</b>			
<b>If no action is taken or required, please say why</b>			
<b>Any other comments</b>			

## **Appendix 7: Interview schedule**

### **Interview Schedule: An exploration of how parents manage their child's cancer-related pain at home**

#### Ground rules

- Some people find research interviews like this one to be helpful. Some people find these upsetting. We may cover topics which are difficult to talk about. If you do become upset, please remember:
  - o This interview is entirely voluntary.
  - o You can stop at any point. You can pause at any point.
  - o You do not have to give a reason for withdrawing. Your child's care will not be affected whether you participate or decide to withdraw.
  - o There are support services available via your child's healthcare team, psychology services and charities like CLIC Sargent.
- This interview is confidential – this means that:
  - o After this interview I will remove identifiable information from the transcript before I share it with the research team.
  - o I might choose to publish some of the quotes from this interview which means you will be able to identify yourself, but others will not be able to identify you.
  - o I will not share what you have said in this interview with your doctors or your healthcare team.
  - o If you say something to me which makes me think that you, or someone else's safety is at risk, I may have to break your confidentiality. I would normally discuss this with you before doing this.
- I will be recording the interview so that I don't forget what is said, I may also occasionally make some notes.
- Are you still happy to go ahead?

#### Introductory questions

- Can you tell me a little bit about yourself, \*child's name\*, and your journey through cancer?
- With focus on pain at home, can you tell me how pain has affected \*child's name\* through their cancer journey?
- Probing questions:
  - o Can you tell me how \*child's name\* pain has affected you?
  - o Can you tell me how \*child's name\* pain has affected your family?

### Subjective norms

- Can you tell me about the people who support you in managing \*child's name\* pain?
- Probing questions:
  - o Are you sufficiently supported?
  - o Do you agree / disagree with the advice these people give you?
  - o How has your support changed?

### Perceived behavioural control

### Internal factors

- Can you tell me about how confident do you feel in managing \*child's name\* pain?
- Probing questions:
  - o How has this changed? Why has this changed?
  - o How do you know \*child's name\* is in pain? How do you know what to do?

### External factors

### Facilitators

- Can you tell me about things which help you manage \*child's name\* pain?
- Probing questions:
  - o Are there any helpful tips and tricks you've picked up?
  - o Are there things you wish you'd know with hindsight?

### Barriers

- Can you tell me about things which prevent you managing \*child's name\* pain?
- Probing questions:
  - o What have been the biggest challenges to managing \*child's name\* pain?

### Closing questions

- If you were giving one piece of advice to another parent about managing their child's cancer related pain at home what would that be?
- Is there anything else you'd like to say?
- \*Researcher to give a summary of the discussion\*
- How do you think this interview and research has affected you?
- Would you like to receive a summary of the results?
- Thank you for your time.

**Appendix 8: Consent form - interview**

# An exploration of how parents manage their child's cancer-related pain at home

**Consent form – Interview – Participant copy**

To indicate consent,  
 please initial in the box

I have read and been given a copy of the participant information sheet (Version 1, 26/09/16) which describes the research in which I have been asked to participate.

The researcher has explained the nature and purpose of the research and I understand what is being proposed.

I have been given the opportunity to ask questions about the study and have received answers to my satisfaction.

I understand that taking part in the study will not affect the care of my child.

I understand that taking part is voluntary and that I may withdraw at any time and do not have to give a reason for withdrawing.

I have been informed about, and consent to, the interview being audio recorded.

I have been informed about what the data collected will be used for, to whom it may be disclosed, and how long it will be retained.

I hereby consent to participate in the study that has been fully explained to me.


-----

Name	Date	Signature
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As the Researcher responsible for this study I confirm that I have explained to the participant named above the nature and purpose of the research to be undertaken.

-----

Researcher	Date	Signature
------------	------	-----------

If you are at all concerned about this study, please contact: Roslyn Parker, [parker11@lsbu.ac.uk](mailto:parker11@lsbu.ac.uk)

# An exploration of how parents manage their child's cancer-related pain at home

**Consent form – Interview – Researcher copy**

To indicate consent,  
please initial in the box

I have read and been given a copy of the participant information sheet (Version 1, 26/09/16) which describes the research in which I have been asked to participate.

The researcher has explained the nature and purpose of the research and I believe that I understand what is being proposed.

I have been given the opportunity to ask questions about the study and have received answers to my satisfaction.

I understand that taking part in the study will not affect the care of my child.

I understand that taking part is voluntary and that I may withdraw at any time and do not have to give a reason for withdrawing.

I have been informed about, and consent to, the interview being audio recorded.

I have been informed about what the data collected will be used for, to whom it may be disclosed, and how long it will be retained.

I hereby consent to participate in the study that has been fully explained to me.


-----

Name	Date	Signature
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As the Researcher responsible for this study I confirm that I have explained to the participant named above the nature and purpose of the research to be undertaken.

-----

Researcher	Date	Signature
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If you are at all concerned about this study, please contact: Roslyn Parker, [parker11@lsbu.ac.uk](mailto:parker11@lsbu.ac.uk)

### Questions about you

What is your relationship to your child?	Mother / Father / Other (please describe)
What is your age? (please circle)	<input type="checkbox"/> 18-24 years <input type="checkbox"/> 25-34 years <input type="checkbox"/> 35-44 years <input type="checkbox"/> 45-54 years <input type="checkbox"/> 55-64 years <input type="checkbox"/> 65 years or older
Please tick one option which best describes your ethnic group or background:	<input type="checkbox"/> White <input type="checkbox"/> Mixed / Multiple ethnic groups <input type="checkbox"/> Asian / Asian British <input type="checkbox"/> Black / African / Caribbean / Black British <input type="checkbox"/> Other ethnic group (please describe) _____
What is your annual household income? (please tick)	<input type="checkbox"/> Less than £14,000 per year <input type="checkbox"/> £15,000 – £24,000 per year <input type="checkbox"/> £25,000 – £39,000 per year <input type="checkbox"/> £40,000 – £59,000 per year <input type="checkbox"/> More than £60,000 per year
Please tick which best describes your educational level:	<input type="checkbox"/> Didn't finish school <input type="checkbox"/> Finished school <input type="checkbox"/> Certificate or partial studies at college/universities <input type="checkbox"/> Completed a bachelor's degree <input type="checkbox"/> Completed a postgraduate degree
Do you have health-related education? (please circle)	Yes / No
If yes, please describe:	

### Questions about your child

How old is your child?	
What is your child's gender?	Male / Female (please circle)
Please circle one option which best describes your child's ethnic group or background:	<ul style="list-style-type: none"> <li>• White</li> <li>• Mixed / Multiple ethnic groups</li> <li>• Asian / Asian British</li> <li>• Black / African / Caribbean / Black British</li> <li>• Other ethnic group (please describe) _____</li> </ul>
What is your child's diagnosis?	
How long ago was your child diagnosed?	
Which hospitals are involved in your child's cancer care?	



## Appendix 9: Health Research Authority - ethics approval



Health Research Authority

Professor Theresa Wiseman  
The Royal Marsden NHS Foundation Trust,  
Fulham Road,  
London,  
SW3 6JJ

Email: [hra.approval@nhs.net](mailto:hra.approval@nhs.net)

21 November 2016

Dear Professor Wiseman

### Letter of **HRA Approval**

<b>Study title:</b>	<b>An exploration of how parents manage their child's cancer-related pain at home</b>
<b>IRAS project ID:</b>	<b>203527</b>
<b>Protocol number:</b>	<b>CCR4569</b>
<b>REC reference:</b>	<b>16/NS/0121</b>
<b>Sponsor</b>	<b>The Royal Marsden NHS Foundation Trust</b>

I am pleased to confirm that **HRA Approval** has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications noted in this letter.

#### **Participation of NHS Organisations in England**

The sponsor should now provide a copy of this letter to all participating NHS organisations in England.

*Appendix B* provides important information for sponsors and participating NHS organisations in England for arranging and confirming capacity and capability. **Please read *Appendix B* carefully**, in particular the following sections:

- *Participating NHS organisations in England* – this clarifies the types of participating organisations in the study and whether or not all organisations will be undertaking the same activities
- *Confirmation of capacity and capability* - this confirms whether or not each type of participating NHS organisation in England is expected to give formal confirmation of capacity and capability. Where formal confirmation is not expected, the section also provides details on the time limit given to participating organisations to opt out of the study, or request additional time, before their participation is assumed.
- *Allocation of responsibilities and rights are agreed and documented (4.1 of HRA assessment criteria)* - this provides detail on the form of agreement to be used in the study to confirm capacity and capability, where applicable.

Further information on funding, HR processes, and compliance with HRA criteria and standards is also provided.

It is critical that you involve both the research management function (e.g. R&D office) supporting each organisation and the local research team (where there is one) in setting up your study. Contact details and further information about working with the research management function for each organisation can be accessed from [www.hra.nhs.uk/hra-approval](http://www.hra.nhs.uk/hra-approval).

### Appendices

The HRA Approval letter contains the following appendices:

- A – List of documents reviewed during HRA assessment
- B – Summary of HRA assessment

### After HRA Approval

The document “*After Ethical Review – guidance for sponsors and investigators*”, issued with your REC favourable opinion, gives detailed guidance on reporting expectations for studies, including:

- Registration of research
- Notifying amendments
- Notifying the end of the study

The HRA website also provides guidance on these topics, and is updated in the light of changes in reporting expectations or procedures.

In addition to the guidance in the above, please note the following:

- HRA Approval applies for the duration of your REC favourable opinion, unless otherwise notified in writing by the HRA.
- Substantial amendments should be submitted directly to the Research Ethics Committee, as detailed in the *After Ethical Review* document. Non-substantial amendments should be submitted for review by the HRA using the form provided on the [HRA website](http://www.hra.nhs.uk), and emailed to [hra.amendments@nhs.net](mailto:hra.amendments@nhs.net).
- The HRA will categorise amendments (substantial and non-substantial) and issue confirmation of continued HRA Approval. Further details can be found on the [HRA website](http://www.hra.nhs.uk).

### Scope

HRA Approval provides an approval for research involving patients or staff in NHS organisations in England.

If your study involves NHS organisations in other countries in the UK, please contact the relevant national coordinating functions for support and advice. Further information can be found at <http://www.hra.nhs.uk/resources/applying-for-reviews/nhs-hsc-rd-review/>.

If there are participating non-NHS organisations, local agreement should be obtained in accordance with the procedures of the local participating non-NHS organisation.

### User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application

IRAS project ID	203527
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procedure. If you wish to make your views known please email the HRA at [hra.approval@nhs.net](mailto:hra.approval@nhs.net). Additionally, one of our staff would be happy to call and discuss your experience of HRA Approval.

#### **HRA Training**

We are pleased to welcome researchers and research management staff at our training days – see details at <http://www.hra.nhs.uk/hra-training/>

Your IRAS project ID is **203527**. Please quote this on all correspondence.

Yours sincerely

**Miss Lauren Allen**

**Assessor**

Email: [hra.approval@nhs.net](mailto:hra.approval@nhs.net)

Copy to: *Julie Curtis, The Royal Marsden NHS Foundation Trust (Sponsor contact & Lead NHS R&D contact)*

## Appendix 10: The Royal Marsden NHS Foundation Trust - ethics approval

### The ROYAL MARSDEN NHS Foundation Trust

Chairman: Dr Alistair Ring  
R&D-CCR@rmh.nhs.uk

Research & Development Office  
West Wing, Downs Road  
RM Sutton  
Tel: 020 8661 3871

Professor Theresa Wiseman  
Strategic Lead for Health Service Research  
Royal Marsden NHS Foundation Trust  
Fulham Road  
London  
SW3 6JJ

14<sup>th</sup> November 2016

Dear Professor Wiseman,

**CCR4569 An exploration of how parents manage their child's cancer-related pain at home**  
**IRAS Number: 203527**

Further to scientific review by the Committee for Clinical Research (CCR) I am pleased to advise you that The Royal Marsden NHS Foundation Trust has accepted the role of sponsor for the above study and will undertake the responsibilities of research sponsor, in accordance with the Research Governance Framework 2005 (2<sup>nd</sup> edition) subject to compliance with the following and in accordance with the relevant Trust policies and Standard Operating Procedures (SOPs):

**1.0 Sponsor Approval**

The study must be approved by the Sponsor and at the Royal Marsden NHS Foundation Trust this means receipt of the CCR Sponsor approval email. Patient recruitment to the study must not commence until all regulatory approvals are in place.

**1.1 Research Ethics Committee (REC) Favourable Opinion**

Where applicable the study must be approved by REC and it is a requirement to abide by the conditions set out in the letter of favourable opinion. This approval letter must then be forwarded to R&D.

**1.2 Health Research Authority (HRA) Approval**

To ensure that the study has been approved by HRA and to abide by the conditions set out in the approval letter.

**1.3 Research & Development Confirmation of Capability and Capacity**

To ensure the study has received local confirmation of capability and capacity from R&D (including relevant support services) at participating sites. This must be in place prior to the site opening.

**2.0 Research Governance**

All members of the research team must work in accordance with the set of standards outlined in the Department of Health's Research Governance Framework for Health and Social Care second edition issued in April 2005. This is available at [https://www.gov.uk/government/uploads/system/uploads/attachment\\_data/file/139565/dh\\_4122427.pdf](https://www.gov.uk/government/uploads/system/uploads/attachment_data/file/139565/dh_4122427.pdf)

Studies involving Investigational Medicinal Products (IMPs) must be conducted in accordance with the Medicines for Human Use (Clinical Trials) Regulations 2004 available at

<http://www.opsi.gov.uk/si/si2004/20041031.htm> and The Medicines for Human Use (Clinical Trials) Amendment Regulations 2006 available at <http://www.opsi.gov.uk/si/si2006/20061928.htm> and as amended from time to time.

Studies involving Medical Devices (IMDs) must be conducted in accordance with the Medical Devices Regulations 2002 available at <https://www.gov.uk/topic/medicines-medical-devices-blood/medical-devices-regulation-safety>.

Where tissue samples are involved, the study must be carried out in accordance with the Human Tissue Act 2004 available at <http://www.legislation.gov.uk/ukpga/2004/30/contents> and the Codes of Practice issued by the Human Tissue Authority available at [http://www.hta.gov.uk/guidance/codes\\_of\\_practice.cfm](http://www.hta.gov.uk/guidance/codes_of_practice.cfm). Where tissue is to be transferred from the Royal Marsden to an external organisation an appropriate agreement should be in place.

**2.1 Good Clinical Practice (GCP)**

All research staff participating in this study are required to attend GCP training to ensure they understand their legal requirements, delegated duties and responsibilities to conduct the trial according to protocol. It is the Chief Investigator's responsibility to ensure that staff are qualified and adequately trained to carry out the duties they have been delegated, as outlined in the delegations log.

**2.2 Protocol**

The protocol has been scientifically reviewed and approved by a Research Ethics Committee and Regulatory Authorities. All staff must conduct the trial according to the procedures outlined in the approved protocol and details as outlined in the relevant signed agreement.

**2.3 Delegation of Responsibilities**

The Royal Marsden NHS Foundation Trust has accepted the role of sponsor based on the allocation of responsibilities detailed in Appendix 1. All staff participating in the study are required to undertake all responsibilities allocated to them. The Chief Investigator is responsible for ensuring staff involved in the study know and understand their responsibilities.

**2.4 Research agreements**

Under the Research Governance Framework, 2<sup>nd</sup> edition, 2005 and the Medicines for Human Use (Clinical Trials) Regulations 2004 and Amendment Regulations 2006, research agreements must be in place with all other participant organisations. For multicentre trials fully executed copies of the site agreements should be held in the trial master file. Please make sure that you have a list of all participating centres, together with all regulatory approvals including R&D confirmation of capability and capacity from all sites.

**2.5 NHS Indemnity**

The NHS Litigation Authority will cover standard clinical negligence by employees, staff and health professionals employed by the Royal Marsden through the Clinical Negligence Scheme for Trusts.

More information is available at <http://www.nhs.uk/Claims/Pages/Clinical.aspx>. There is unlimited liability and no excess. Insurance is provided under the Clinical Negligence Scheme for Trusts and there is no cover for non-negligence claims.

For all notification of claims please contact the Board Secretariat.

All collaborating centres are responsible for ensuring they have their own insurance and indemnity arrangements for the duration of the research.

**2.6 Audit / Inspection**

The Chief Investigator will ensure that all trial documentation is available for inspection by regulatory authorities and or other auditors as required. This includes the Trial Master File, source documentation such as patients' medical records, case report forms, lab reports, lab notes, etc.

- 3.0 Protocol violation or Breaches in GCP**  
Where the safety of a patient or the validity of study data are at risk because of a breach in protocol this should be notified to the R&D immediately.
- 3.1 Staffing issues**  
Where there are issues affecting the patient recruitment, trial resources and or capacity to run a trial due to the lack of staff, the R&D must be notified as soon as the research team become aware of this.
- 4.0 Safety Reporting**  
You are reminded that all serious adverse events (SAEs) must be identified, collected and recorded in case report forms and in accordance to the study protocol. The expectedness, causality and severity of all AEs must be documented in accordance to the study protocol. All Serious Adverse Events should be reported to the main REC and Clinical R&D within the 15 day timeframe.
- Please be aware of the Trust policy: *Accident/Incident Reporting and Investigation Policy Including Serious Untoward Incidents (482)*
- 5.0 Annual Progress Reporting**  
There is a legal requirement to submit annual progress monitoring reports to the main REC on the anniversary of favourable ethical opinion and copies sent to R&D.
- 5.1 Notification of change in study status**  
R&D must be notified if there is a change in study status to abandoned, on hold, suspended, closed to recruitment, on long-term follow-up or closed through completion. There is a template notification form available on the RM/ICR intranet. For notification of premature closure of the study or completion of a study the *Notification of End of Trial* form should be submitted to main REC and a copy sent to R&D.
- 5.2 Study Amendments**  
Where there is a change in trial arrangement, procedure or conduct of the trial these should be notified to R&D as a substantial or non-substantial amendment using the relevant IRAS form available on their website. All amendments must be sent through to the R&D Office with the internal amendment cover sheet for review and approval by the Chair or Acting Chair of the Committee for Clinical Research (CCR). As soon as the amendment has been approved by CCR and REC/HRA, R&D confirmation will be given.
- 6.0 Informed Consent**  
It is a legal requirement to ensure that all patients have been provided the necessary information to obtain documented informed consent in order to participate in the study according to the protocol inclusion criteria. The signed original consent form should be kept in the Trial Master File and copies in the medical notes and a copy given to the patient.
- Please ensure that you abide by the consenting requirements of the Trust's *Consent to Examination or Treatment Policy (325)* available on the RMH intranet.
- 6.1 Recording patient recruitment on the Hospital Information System (HIS)**  
Where Royal Marsden is a site, patients must not be recruited to the trial until you have received notification of HIS activation. This is outlined in your R&D confirmation of capability and capacity email. You are reminded that for all studies, patient accrual at Royal Marsden must be recorded on the "CCR PAT" section of the Hospital Information System.
- 7.0 Maintenance of the Study master file**  
The Chief Investigator is responsible for maintaining the study master file. This should include signed consent forms for Royal Marsden patients which should be stored in a secure location. Signed consent forms for non-Royal Marsden patients should be maintained at the individual participating sites involved in the study. The transfer of ownership of data or of documentation

should be documented. The Chief Investigator will ensure that the contents of trial master file will be archived in accordance with local procedures.

7.1 **Medical Records**

The Chief Investigator should ensure that medical files of trial subjects are retained for at least five years after the conclusion of the trial or in accordance with local procedures.

7.2 **Archiving**

Trial Master File and related study source data documentation (questionnaires, Case Report Forms, lab notes, lab reports, images, medical notes, data collected on PACS or EPS) need to be retained in accordance with local procedures.

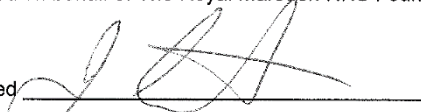
The Chief Investigator will nominate an individual responsible for archiving contents of the trial master file and ensure that access to the archived files should be restricted to these named individuals. The transfer of this responsibility should be documented.

**THE SPONSOR RESERVES THE RIGHT TO SUSPEND THE STUDY IF NOT COMPLIANT**

The Clinical R&D Office can provide support and advice to you and your team in the conduct of the study. Please do not hesitate to contact Research & Development on **020 8661 3871** or [research.development@rmh.nhs.uk](mailto:research.development@rmh.nhs.uk) or **fax: 020 8915 6700**.

Sponsorship has been granted for the above study titled **CCR4569 An exploration of how parents manage their child's cancer-related pain at home**

Signed on behalf of The Royal Marsden NHS Foundation Trust

Signed 

**Julie Curtis**  
Clinical R&D Manager

Date: 14th November 2016.



## Appendix 11: London South Bank University - ethics approval



E-mail: [ethics@lsbu.ac.uk](mailto:ethics@lsbu.ac.uk)  
UEP Ref No: **UEP1116**

24<sup>th</sup> November 2016

Dear Roses Parker,

**RE: 'An exploration of how parents manage their child's cancer-related pain at home'.**

Thank you for submitting your documentation and approval letters from HRA, NHS REC and your local R&D office.

I am writing to inform you that ethical approval for your proposal has been upheld by the LSBU University Ethics Panel by Chair's action (by Professor Shushma Patel).

We wish you every success with your research.

Yours sincerely,



Ms. O. Imonioro  
Compliance and Systems Manager – on behalf the LSBU University Ethics Panel

cc:  
Prof Shushma Patel, Chair, LSBU University Ethics Panel

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**Appendix 12: Shapiro-Wilk test to ascertain distribution of scales and sub-scales**

Scales / sub-scales	Shapiro-Wilk Statistic	df	Sig.
PPEP	.949	33	.12
PPEP active loud	.922	33	.21
PPEP quiet inactive	.953	33	.16
PPEP attention seeking	.920	33	.18
MAQ	.984	33	.90
MAQ avoidance	.926	33	.27
MAQ appropriate use	.979	33	.751
MAQ fear of side-effect	.972	33	.53

The test rejects the hypothesis of normality when the p-value is less than or equal to 0.05

**Appendix 13: Combined agree, disagree, and unsure responses for PPEP and MAQ items compared with previous research**

Author, year	Parker 2017			Zisk 2010			Zisk 2007			Fortier 2012			Twycross 2015			Sub-scale
Clinical condition	Cancer			Surgery			Surgery			Cancer			General population			
Country	UK			US			US			US			UK			
Item	Disagree	Unsure	Agree	Disagree	Unsure	Agree	Disagree	Unsure	Agree	Disagree	Unsure	Agree	Disagree	Unsure	Agree	
Children should be given pain medication as little as possible because of side-effects	37	24	38	52	6	42	36	13	51	48	10	42	49	17	34	Avoidance
Children who take pain medication for pain may learn to take drugs to solve other problems	43	23	34	75	12	13	66	19	15	63	17	21	74	12	14	Avoidance
Pain medication works the same no matter how often it is used	42	24	34	77	16	7	73	16	12	69	18	13	60	18	22	Fear of side-effects
Pain medication works best when it is given as little as possible	40	22	38	53	24	24	53	19	28	42	17	41	44	19	37	Avoidance
Pain medication has many side-effects	21	27	52	39	24	38	33	37	30	18	29	54	42	22	36	Fear of side-effects
Children will become addicted to pain medication if they take it for pain	46	21	33	82	9	9	72	22	6	59	20	21	75	13	12	Avoidance

Author, year	Parker 2017			Zisk 2010			Zisk 2007			Fortier 2012			Twycross 2015			Sub-scale
There is little need to worry about side-effects from pain medication*	49	18	33	57	14	29	62	18	20	63	20	17	34	28	38	Fear of side-effects
It is unlikely a child will become addicted to pain medication if taken for pain*	17	33	50	13	20	68	17	23	60	26	27	47	61	17	22	Appropriate use
Pain medication is addictive	29	28	42	21	27	52	37	20	43	20	17	63	50	22	28	Fear of side-effects
Pain medication works best if saved for when the pain is quite bad	43	9	48	62	9	29	66	12	22	38	8	54	40	13	47	Avoidance
Using pain medication for children's pain leads to later drug abuse	55	17	28	86	10	4	80	18	2	70	21	9	84	9	8	Avoidance
There is little risk of addiction when pain medication is given for pain*	21	18	60	22	18	60	19	22	59	30	23	47	21	18	61	Appropriate use
Children learn how to use pain medication responsibly when it is given for pain*	10	27	63	8	14	78	4	28	69	12	19	69	14	24	62	Appropriate use
Side-effects are something to worry about when giving children pain medication	16	11	73	17	10	73	17	10	73	14	10	76	31	22	47	Fear of side-effects

Author, year	Parker 2017			Zisk 2010			Zisk 2007			Fortier 2012			Twycross 2015			Sub-scale
The less often children take pain medication for pain, the better the medicine works	37	25	37	48	15	37	39	24	37	39	24	38	37	15	48	Avoidance
Giving children pain medication for pain teaches proper use of drugs*	21	33	46	25	21	55	19	22	59	14	29	57	28	21	51	Appropriate use

\*Reverse scored item

**Appendix 14: Causes of pain at home as reported by parents**

This table presents a list of causes of pain as reported by parents of children with cancer in pain diaries. Every cause listed has been represented. Causes have been grouped to aid the reader. Duplicates were merged so frequency of each cause is not represented here.

Drug related	Constipation	Inactivity
Chemotherapy	Diarrhoea	Muscle weakness
Chemotherapy in stools causing bottom pain	Food	Weakness
Codeine side-effect	Hunger	Everyday life related
Cytarabine	Lack of fluids	Being active
Doxorubicin	Nausea	Environment
Drug side-effects	Needing to go to toilet	Heavy school bag
Irinotecan	Reduced appetite	Pulled muscle
Jaw ache	Sickness	Teething
Laxatives	Stomach cramps	Too much screen time
Medication	Vomiting	Tumour on spine was compressed on a ride
Oral chemo	Disease	Psychological
Pegaspargase	Disease	Anxiety
Pregabalin side-effects	Procedure related	Imagination
Steroids	Bone Marrow Aspirates	Pain descriptor
Vincristine	General anaesthetic	Headache
Comorbid condition	Injection	Joint pain
Avascular Necrosis	Intrathecal	Soreness
Cold	Lumber puncture	Miscellaneous
Conjunctivitis	Lung function test	Combination
Cough	MRI scanner table surface (hard surface) aggravated his back	Dizziness
Fever	Port access	Dryness
Hand / foot syndrome	Port de-access	Other
Infection	Procedures	Pins and needles
Low blood count	Removal of Hickman line dressing	Rash
Virus	Surgery	Tiredness
VOD	Surgical removal of impacted eyeteeth under GA	Don't know
Mucositis	Movement related	Don't know
Mouth ulcers	Exercises	
Mucositis		
Bowel or gut related		
Anal tear		
Bowels		