‘Quality of life’ in children with high-risk brain tumours: children’s, parents’ and healthcare professionals’ perspectives over the course of the illness.

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Declaration

I, Emma Beecham, confirm that the work presented in this thesis is my own. Where information has been derived from other sources, I confirm that this has been indicated in the thesis.

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ABSTRACT

BACKGROUND: Children with high-risk brain tumours (HRBTs) and their families face many decisions in which quality of life (QoL) is a factor. Previous QoL research has focused on quantitative measurement, leaving a gap as to what QoL means from a child's, parent's and clinician's perspective.

Ethical guidance and institutional policies call for children to participate in these decisions as appropriate, yet there is little recognition of children's unique modes of participation.

AIMS: To provide a robust description and understanding of QoL as it emerges during consultations from the perspective of the child with an HRBT, the parents and clinicians, and across the illness trajectory.

METHODS: Mixed qualitative and interactionist methods were employed. QoL dimensions were developed to explore QoL. Thematic analysis and constant comparison were used to reveal similarities and differences between the parents' (N=24) and clinicians' (N=14) QoL perspectives. Using the case of a 14-year-old female and taking a fresh approach to participation, including non-verbal language, discourse and conversation analysis were used to uncover the child's voice in QoL discussions.

RESULTS: The importance of key dimensions of QoL changed for parents and clinicians over the course of the illness as did their ideas about the child's future and of a normal life. Parents' and clinicians' viewpoints diverged as the illness progressed. The child's voice was co-constructed together with the clinician and the parents. Child-centred behaviours on the part of clinicians and parents facilitated this co-construction.

CONCLUSIONS: The multi-dimensional, subjective and dynamic nature of QoL, pose challenges for the conceptualisation of QoL. Elicitation of children's own views of their QoL requires an analysis of the triadic interactions in which their views emerge. The results of these analyses provide a foundation for design of education and training for clinicians to manage these complex interactions.

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IMPACT STATEMENT

The current understanding of what constitutes Quality of Life (QoL) in children with high-risk brain tumours (HRBTs) is based largely on the formulation and the application of quantitative measures. This approach ignores children's, parents' and treating clinicians' views of the QoL of these ill children.

Applying qualitative and interactionist methods to on-the-ground clinical consultations, this thesis sheds light on the triadic interaction (between the child, parent[s], and clinician) in paediatric consultations involving children with BTs. Thematic analysis and constant comparison found that parents' and clinicians' QoL concerns were subjective, dynamic, and temporal, posing challenges for those who rely primarily or exclusively on static cross-sectional QoL instruments. These findings contribute to the conceptual underpinning and theory of QoL.

Looking beyond academia, these results have implications for clinical practice and education and policy. Social science research on communication is not reaching physicians or clinical practice (Gulbrandsen et al, 2022). Gulbrandsen and colleagues highlight the importance of understanding the "physiology of interaction" as described by social scientists to improve clinicians' communication skills. The results from this thesis can directly inform the revision of medical communication training in two ways:

- Firstly, support for clinicians to be cognisant of where the child is in the illness trajectory and recognise key moments at which a parent and clinician's viewpoints on QoL may diverge. This awareness can help avoid the misalignment of views or conflict between clinicians and parents that has been shown to lead to relationship breakdowns.
- Secondly, being aware of how parents and clinicians can facilitate co-construction through child-centred behaviours can aid in the conduct of the paediatric consultation. Implementing these findings on child participation and developing communication training for clinicians could help support and enhance children's participation in the consultation, resulting in a transmission of social science to the bedside.

Ethical guidance and institutional policies urge children's involvement in decision-making (Royal College of Paediatrics and Child Health, 2000; American Academy of Pediatrics Committee on Bioethics, 1995). However, there is little clarity on achieving this. Discourse analysis revealed that the child participated in the QoL conversations in a myriad of ways that are not always verbal and direct. The clinicians' and parents' co-construction of the child's views alongside the child's suggests the need to reframe recommendations for involving the child to include attention to the family members' role in the interaction in the consultation rather than focusing on the child's verbal utterances alone.

Dissemination of this research and reaching out to practice has been a constant mission throughout this PhD programme, starting with the open access publication of a systematic review on BT survivors' concept of QoL (Beecham et al, 2019). Ten national and international presentations on the thesis have been given (see Appendix 1), with empirical findings presented directly to the paediatric palliative care team from the study paediatric hospital. Future plans include writing up two papers from the empirical study which will be further disseminated through social media platforms including Twitter, LinkedIn, and podcasts linked to journals with the aim of also reaching the families themselves.

ABBREVIATIONS KEY

Abbreviation	Full term/Phrase
CNS	Central nervous system
CA	Conversation analysis
DA	Discourse analysis
Dx	Diagnosis
EoL	End of life
HCP	Healthcare professional
HRBT	High-risk brain tumour
PC	Palliative care
POC	Place of Care
QoL	Quality of life
Tx	Treatment
UK	United Kingdom

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PREFACE

‘Alia Singh’ sits with her mother and father in the waiting room of a large tertiary paediatric cancer centre waiting to be seen by a neuro-oncologist. Barely a teenager, she and her parents have come thousands of miles looking for possible further treatment after yet another recurrence of her brain tumour, after three years of treatment marked by chemotherapy, radiation, and surgeries; not to mention hair loss, weight loss, gastrostomy feeds, and cognitive insults. What will Alia and her parents be told? What will they be offered? What will they decide to do? How will they make the decision?

This scenario, like others in this thesis, unfolds over the course of multiple consultations with multiple professionals in the clinic, hospital, and homes over the course of the child’s care and treatment for a high-risk brain tumour, where median survival is less than two years. The reader enters these consultations through an examination of the verbatim transcripts of the audio-recorded consultations. Using principles and practices from conversation analysis and discourse analysis as well as constant comparison, the spoken and unspoken realities in which patients, parents and clinicians find themselves come to light. Particular attention is given to discussions of quality of life, and how the meaning changes for patients, parents and professionals over the course of the illness. In this thesis all voices are heard and examined as patients, parents, and clinicians speak among themselves; offering an opportunity to both observe participation and, not inconsequentially, to see how participation is achieved.

CHAPTER 1 – INTRODUCTION

1.0 CHAPTER OVERVIEW

This chapter introduces the thesis by first discussing the research problem and the gap in the literature to which it is addressed. It summarises the aims and objectives and explains the significance and the academic and clinical contributions of the work. The chapter also includes a summary statement for each of the chapters. For a glossary of the terms used in this thesis see Appendix 2.

1.1 – CONTEXT AND BACKGROUND TO THE THESIS

Brain Tumours (BT) occur in children and young people at all points from infancy through to young adulthood. They fall on a spectrum from slow-growing and benign to those that are highly malignant with a poor prognosis. BTs are still the leading cause of cancer-related death in childhood (Pollack, 1994; Subramanian & Ahmed, 2021). Many children, however, now survive longer, with 75% of them surviving for more than five years (Jones et al, 2018). Even the revolution in molecular biology, providing insights into the genetic basis for most types of childhood BT, has not led to significant advances in the treatment (Tx) of HRBTs such as diffuse intrinsic pontine gliomas (DIPG) (Hayden et al, 2021). Most children with these more aggressive tumours will not achieve long-term survival. As has been the case generally with cancer (Wiener and Bluebond-Langner, 2019), Tx progress has led to a widening of the scope of disease-related issues to which clinicians and researchers attend. Numerous studies have found that those children who survive suffer from many late effects (side effects that occur months or years after the Tx has ended) from the Tx they had received. As a result, clinicians' concerns have moved from a focus on survival alone to examination of the *quality* of that survival (Spieth & Harris, 1996).

The interest in quality of survival has led to efforts to measure quality of life (QoL) in BT survivors. Existing instruments measuring QoL in multiple populations have been adapted and the development of new ones pursued. These scales have been employed to

evaluate the impact of interventions and Txs on QoL and the use of QoL instruments in clinical trials of Txs for BTs has been strongly recommended. The focus on QoL in children with BTs has thus been about an objective, quantifiable property within a defined population.

My recent systematic review of the understanding of QoL in children with BTs, their parents and their clinicians, revealed not only the paucity of research on the idea of QoL among these participant groups (Beecham et al, 2019; see Appendix 3) but also and more specifically, the issues surrounding its meaning to and use by participants in clinical consultations. The review, which revealed the perspectives of young adult BT survivors, concluded that there is a need to understand the concept from all three perspectives, and how the understanding of QoL may vary across the illness trajectory and according to the context in which it is used (e.g. consultations about Tx options, place of care, family relationships).

This thesis aims to fill some of the gaps in the understanding of the crucial subjective side of QoL revealed by the review. It also takes research on QoL in children with BTs into a virtually unexplored area: QoL in Tx decisions when cure is not the goal, when Txs, though perhaps disease-directed in nature, are given to extend life but are not able to accomplish a cure. The first aim of the thesis was chosen due to the systematic review revealing a gap specifically in the perspectives of children with HRBTs who are undergoing Tx many of whom will not survive, their parents and the clinicians in charge of their care.

The examination of child, parent and clinician perspectives of QoL is accomplished through analysis of verbatim transcripts of consultations with paediatric oncologists in the hospital and with the palliative care team in the home. The transcripts come from a wider ethnographic study titled ‘Understanding decision-making for children with high-risk brain tumours (HRBT): A prospective, longitudinal study of parents, children and clinicians to provide guidance for clinical consultations’ (referred to from here on in as the ‘HRBT project’) (Bluebond-Langner et al, 2013a). The HRBT project commenced January 2014 and is ongoing. Data collection was conducted between February 2014 – October 2015 at a joint university hospital and a tertiary paediatric hospital. The project focused on

interactions as they took place in real-time, amongst clinicians, parents and children with HRBTs from diagnosis through to the death of the child or the end of the 20-month data collection period (see Appendix 4 for the HRBT project one-page project overview). This PhD project, not part of the originally funded HRBT project proposal was developed independently. The original HRBT project was funded by the Health Foundation to explore decision making. I was working in a link Research Fellow post between the Louis Dundas Centre for Children's Palliative Care and the Marie Curie Palliative Care Department (MCPCD), and the MCPCD funded my PhD work. Additional funding was also granted by the Health Foundation for further use of the HRBT project's extensive database for other purposes. This included index and attribute coding of the consultation and home visit transcripts, of which I was involved in my Research Fellow role, and my PhD benefited from this coding having already been completed. This PhD started in February 2016, four months after completion of the HRBT project's data collection.

The need to capture the child's perspective and the limited children's data retrieved from the initial analysis, using only verbalisations, led to the second aim of the thesis where the child's voice was explored in further depth using different methods. To understand the child's perspective on QoL dimensions, first it was necessary to examine how children participate in the consultation to understand how their voices emerge in discussions pertaining to QoL-related issues and concerns.

1.2 – RESEARCH PROBLEM

Most research undertaken in the area of QoL has focused on the quantitative measurement of QoL. This narrow focus has left a gap in the academic and clinical knowledge base with regard to understanding the actual meaning of QoL from the perspective of the child, parent, and HCP. Children with BTs and their families face a number of decisions throughout their care and Tx in which QoL is a frequently featured factor. These decisions are particularly important in children with HRBTs, where many diagnoses have a poor prognosis, and in some cases, phase I/II clinical trials and experimental therapies are available.

The importance of appropriately including the child in discussions about their own care and Tx has long been recognised (Royal College of Paediatrics and Child Health, 2000; American Academy of Pediatrics Committee on Bioethics, 1995). When QoL is considered, the child's participation is especially important because of the privileged position that an individual holds concerning the assessment of their own QoL. A few previous studies (e.g. Stivers, 2001; Clemente, 2009; 2015) have carried out a close analysis of naturally occurring conversations in paediatric contexts and looked at aspects of the child's participation in consultations.

Stivers (2001) found that the child's participation is the 'product of an interactional negotiation among the physician, the parent, and the child' (p. 277). Stivers also concluded that focusing entirely on who speaks in the consultation may miss the process of this interactional negotiation notably that it actually includes children.

Clemente (2009, 2015) has also explored child participation in consultations qualitatively, using conversation analysis (CA). Clemente (2009, 2015) similarly to Stivers supports a triadic, observational approach to studying child participation, through his studies examining communication strategies in the US and Spain, respectively. Clemente (2009) concluded that the four different child-initiated communication strategies found in the study were supported by the clinicians in the interactions. Clemente highlights that children's participation depends on all participants' actions, not only that of the child, therefore it is important to analyse the actions of all participants in the interaction (Clemente, 2015).

This thesis makes a unique contribution to that literature. It does so by building on the work of Stivers, Clemente and others and by searching for seriously ill children's voices as they are present in real-time conversations about QoL issues in the context of decisions about their care and Tx.

1.3 – AIMS AND OBJECTIVES

i) AIMS OF THE THESIS

1. To provide a robust description and understanding of the QoL-related issues and concerns which arise in consultations about the care and Tx (treatment) of children with a high-risk brain tumour (HRBT). Attention is given to tracking the concerns of children, their parents and healthcare professionals (HCP) involved in their care and Tx as they emerge in consultations over the course of the illness.
2. To examine how children participate in the consultation, with a particular focus on how their voices emerge in discussions pertaining to QoL-related issues and concerns.
3. To consider the implications of the findings from the empirical study for clinical practice, research and policy.

ii) OBJECTIVES OF THE THESIS

1. Development of a method, working both deductively from the literature and inductively through the data, to identify for analysis sections in the consultations in which QoL-related issues are in play.
2. Exploration of how and when QoL-related issues emerge within the consultations for each participant group across all periods of the illness trajectory.
3. Exploration of the child's voice in consultations, using discourse analysis to further understand how children participate in the clinical consultation in discussions pertaining to QoL-related issues and concerns.
4. Development of recommendations for further research, policy and clinical practice.

1.4 SIGNIFICANCE AND JUSTIFICATION OF THE THESIS

This study will contribute to the body of knowledge on the conceptual underpinning of QoL by exploring the views of children, their parents, and clinicians with regard to what QoL means to them. Understanding the perspectives of children with HRBTs, parents and clinicians will in turn aid in the conduct of the consultation, presentation and selection of options of care and Tx. The study will also contribute to the literature on children's participation in the consultation. By exploring the child's voice in discussions where QoL is in play, the thesis will add to the body of knowledge about how to include a child in the consultation and to provide understanding of the child's perspective on their QoL.

1.5 THESIS OUTLINE

In Chapter 1, the goals and contributions of the study have been introduced, including the introduction of the wider HRBT project that provides the data for this thesis. The research aims and objectives have been presented and the value of the research explained. Chapter 2 situates the project in the existing literature, focusing on the measurement of QoL, dimensions of QoL in the literature, and children's participation in clinical consultations. Chapter 3 presents the theoretical perspective and methodological approach which underpins the thesis. Chapter 4 explores the perspectives of parents and clinicians from the HRBT project using a set of 'dimensions of QoL' developed in the thesis and discusses the chapter's findings in relation to previous literature. Chapter 5 explores how the child's voice emerges in the consultation during discussions in which QoL-related issues and concerns are in play and discusses the chapter's findings in relation to the previous literature. Finally, Chapter 6 brings together results from the empirical work in Chapters 4 and 5 and discusses the research and clinical implications of the work, opportunities for future research, and reflections on the methods used.

The following chapter situates the project in the relevant literature.

CHAPTER 2 – SITUATING THE PROJECT

2.0 CHAPTER OVERVIEW

This thesis examines the occurrence and use of QoL concerns and issues in paediatric oncology consultations about the care and Tx of children with HRBTs. How QoL-related concerns emerge in the talk of each of the participant groups in the consultations- children/patients, parents, and clinicians- is examined. Because it is the child's QoL which is in question in these consultations and because QoL is a subjective phenomenon, the child's participation in the consultation is also explored in detail. Therefore, the thesis is situated within two literatures: relevant issues in the QoL literature, and relevant issues in the child participation literature. This chapter addresses relevant aspects of each of these literatures.

2.1 RELEVANT ISSUES IN QUALITY OF LIFE LITERATURE

Before proceeding, a comment on the use of the terms "quality of life" and "health related quality of life" is needed. Much of the literature uses the term health related quality of life and its acronym HRQoL. The purpose is to distinguish their interest in QoL from that of someone interested in learning, for example, where in the UK residents report the best QoL. The term HRQoL is used to indicate a different and perhaps narrower interest in "health-specific" QoL, an interest to which certain desirable features of a location which support a good QoL for residents, for example, are not relevant. Having recognised the distinction, it is noted that this distinction is not of consequence in this thesis. This thesis and all the studies examined throughout the thesis, deal with the concept in the context of health. Some studies use the term QoL, others use HRQoL, and some use both. What is most important to note is that the use of one term or the other does not affect what is said in the thesis about difficulties in measuring QoL nor does it alter the need to define the underlying concept, whether it is called HRQoL or simply QoL.

A. ISSUE OF DEFINITION AND CONCEPTUALISATION

An often neglected but significant discussion in the area of QoL is the issue of the limited conceptual underpinning of QoL and its assessment. Definitions and conceptualisations of QoL widely vary, with implications for the development of QoL measures. A major issue in the field of QoL research is the lack of a clear theoretical underpinning of the concept and theory-driven research (Wallander, 2001; Eiser and Morse, 2001a).

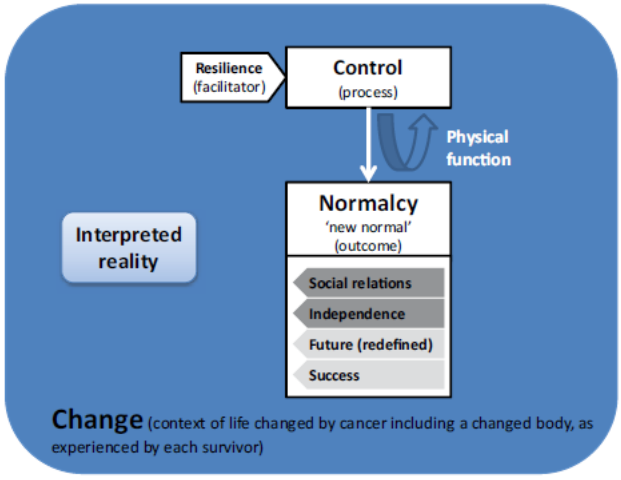
Since the introduction of the term QoL in the 1960's in the medical literature, QoL has become an increasingly popular phrase, finding its way into everyday speech. However, there is still no one universally agreed definition of QoL. In addition, as Haraldstad et al (2019) found in their review on QoL in medicine and health sciences, only 13% of studies on QoL provided a definition.

The World Health Organisation (WHO) originally defined QoL as a 'state of complete physical, mental and social well-being, and not merely the absence of disease and infirmity' (WHO, 1947). However, the WHO has since moved towards a more subjective, nuanced view, defining QoL as 'an individual's perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns' (WHOQOL Group, 1995). Hinds et al (2004), drawing on her research with children with cancer, defined QoL as 'an overall sense of well-being based on being able to participate in usual activities; to interact with others and feel cared about; to cope with uncomfortable physical, emotional, and cognitive reactions; and to find meaning in the illness experience.' Notably, while there appears to be no agreed definition, there is a general consensus among researchers that QoL is a subjective and multidimensional construct (Matza et al, 2004).

More recently, in the context of ill health, QoL has been interpreted conceptually as a reintegration to normal living (Eiser, 2004) and involves striving for Normality, or a 'new normal' (Clarke-Steffen, 1997; Deatrick et al, 1999; Van Schoors et al, 2018; Beecham et al, 2019). My systematic review used a meta-ethnographic exploration to understand previous conceptualisations of QoL in children with BTs. Four studies in the review interviewed young people survivors (three studies used semi-structured interviews and one study used semi-structured interviews and one semi-structured focus group), one

study used semi-structured interviews with children and parents and one study interviewed bereaved parents through a semi-structured focus group. Three studies cited thematic analysis to analyse their data, two cited content analysis and one cited a combination of grounded theory techniques and narrative analysis. The systematic review used the three major strategies of meta-ethnography (Dixon-Woods et al, 2006): reciprocal translational analysis (RTA), refutational synthesis and lines of argument synthesis (LOA) to determine the dimensions of QoL in the concept. Key concepts in each study were identified and then translated into each other, contradictions were characterised and explained and resulting concepts reported (Beecham et al, 2019). The review found that the dimensions involved in the young survivor's QoL concept were: Normalcy, control, independence, social functioning, future, success, physical factors, change and resilience (see Figure 1 below for the Model of QoL from my systematic review) . Overall, the review concluded that a holistic conceptualisation of QoL is 'striving for a new normal' (Beecham et al, 2019). However, the review concluded that these dimensions of the concept were all involved in varying ways and that QoL is not only a multidimensional concept but also multi-faceted i.e. QoL is made up of different dimensions but that these dimensions themselves are nuanced and work in different ways.

FIGURE 1. MODEL OF QOL OF YOUNG PEOPLE SURVIVORS (BEECHAM ET AL, 2019)



In Deatrick et al's (1999) review on the concept of normalisation, authors concluded that change and impairment are acknowledged as part of the concept and that normalisation involves both a cognitive (definitional) and behavioural (strategies) process. Authors also argue that '*No one attribute can be considered without viewing its context and the attributes as a set. While attempting to clarify the conceptual foundation of normalization, defining attributes are necessarily reductionistic'* (p. 213).

Despite this agreement in the literature that a need for normality is what families want and how they understand well-being, measures of QoL do not feature normalcy and achievements. Instead they measure deficits in several domains. As Haase et al (1999) stress, this is a limited way of assessing QoL and without assessing the meaning for the individual child. No measure focuses on how far they can live their normal everyday life. The European Quality of Life-5 Dimensions (EQ5D; The EuroQol Group, 1990) does touch on normality through a domain of how a patient is able to undertake their "usual activities" be it school, study, or work as does the Child Health Utility instrument (CHU9D; Stevens, 2009) with the domain "able to join in activities" but these are one of many domains in the measures.

There is no current gold standard measure for QoL in children with BTs. The Pediatric Quality of Life (PedsQL) scale (Varni et al, 1999), the most frequently used tool to measure QoL in children, has been found to lack relevance to this population of children with life-limiting conditions (Huang et al, 2011).

Lawford and Eiser (2001) suggest that a theoretical model for QoL would clarify the domains and concepts to be assessed in QoL measures (avoiding need for a wide range of current measures involving different domains) and understanding how they are related to one another rather than constituting an unrelated list.

The significance of this discussion for this thesis is twofold. First, what QoL means to people is something that has been and continues to be debated. The thesis recognises that different participants in clinical consultations may approach QoL in different ways. My systematic review revealed the perspectives of young people with BTs who survive, revealing a gap in the literature regarding the perspectives of clinicians, children with a worse prognosis than those in the review, that would not survive the BT, and their parents.

Second, QoL is a complex, multifaceted idea. This viewpoint is demonstrated in the set of dimensions of QoL developed to identify sections of the consultations in which QoL is in play, discussed in Chapter 4.

B. INDIVIDUAL/SUBJECTIVE VS OBJECTIVE ASSESSMENT

Reliance on measures with predetermined domains and fixed weighting of the items and domains can belie the idea that QoL is substantially subjective and implicitly assume that QoL means the same thing to everyone. It is important to understand the child's subjective perspective as previous research has found that parents and HCPs may perceive QoL differently (Janse et al, 2005). Haase et al (1999) argued that items and instruments focused solely on objective health status and functional performance would lead to potentially misleading conclusions about HRQoL. Measures that focus on functional ability assess what individuals *cannot* do rather than what they feel able to do (Eiser and Morse, 2001b). Eiser and Morse (2001b) emphasised the interdependence between QoL and cultural experience and argued that QoL assessment should include both objective and subjective elements. Anthony et al (2017) in their interview study exploring the theoretical underpinnings of QoL concluded:

> '*Our findings suggest that existing PRO [patient reported outcome] instruments contain content that does not reflect the QoL experiences and perspectives of childhood cancer patients and survivors. Further, these measurements weakly address important concerns unique to this population, indicating that the current PRO instruments have limited utility in research and clinical practice*' (p.280).

This discussion cautions us about thinking that some people can have expert knowledge about the QoL of others. Measures are useful for the comparison of the impact of interventions and Txs but may not define what a particular individual's QoL is. This underscores the importance of seeking the child's/patient's views on their own QoL.

C. DYNAMIC NATURE OF QOL

HRQoL is dynamic. Its measurement requires sensitivity to change and differs by location, time, period in the illness and person enquiring/administering the HRQoL measure, to mention but a few considerations supporting the view that the measurement and reporting of QoL are contextual. For example, the same instruments when used at different times and locations have yielded large differences in scores (Hinds, 2010). Similarly, as Ferrans (2004) highlighted the factors influencing an individual's evaluation of their QoL may potentially change over time. This is especially important for the study of children with HRBT where death and dying/end of life (EoL) are prominent and research is lacking (Hinds, 2010). The interactional methods used in this study (see Chapter 3) are well suited to consideration of the context in which HRQoL emerges.

D. ISSUE OF PROXY REPORTS

It has been asked if a certain degree of maturity is necessary to meaningfully report one's QoL. Previous studies have concluded that children as young as five years old can reliably convey their perspective (Bates et al, 1995) and report their own QoL (Varni et al, 2007; Landgraf & Abetz, 1996). Landgraf and Abetz argued, however, that these reports are only reliable on concrete concepts and a more conservative age of 9 or 10 years of age is better if reports of subjective concepts within QoL such as self-esteem are involved. In situations where children are deemed too young to report on QoL, reports of parents are used instead. Children with BTs are often pre-verbal or non-verbal or lack the capacity to express their views. Often referred to as 'parent proxy' response, parents in these cases often then give their views of the child's QoL, which then often stands as the child's view.

A number of issues have been raised repeatedly in conjunction with parent proxy reporting (Johnston et al, 2003). Evidence suggests that there are differences in the evaluation of QoL between chronically ill children, parents, and paediatricians, particularly around pain and emotion (Janse et al, 2005; 2008). Eiser and Morse (2001b) point out that there is greater concordance between child and parent ratings for observable behaviours such as physical functioning than for emotional or social QoL. Although it has been claimed by some that measures have scored well statistically on reliability and

validity tests, there is no accepted gold standard of measure and therefore these claims should be taken with caution. It has been suggested that the preferred reporting source should be the patient (Hinds, 2010). Eiser and Morse (2001b) still see a use for proxy ratings as substitutes or as complementary information about children. They stress that focusing on the concordance or lack of concordance between child and proxy ratings is not helpful and there is value in having both proxy and child reports to provide the extra information that contributes towards a more detailed picture of the child's QoL (Eiser & Morse, 2001a; 2001b). Eiser and Morse (2001a) highlight the importance of proxy reporting not only by parents but also by other significant adults in the children's lives such as clinicians or teachers alongside the need for understanding why differences occur in proxy reporting.

In-depth discourse analysis (DA) of consultations between patient parents and clinicians in Chapter 5 provides another perspective through which the place of proxy reporting when QoL is at issue is considered. In this chapter, attention is given to the child's voice, her participation in the consultations when issues QoL emerge for herself as well as for parents and clinicians.

2.2 RELEVANT ISSUES IN CHILDREN'S PARTICIPATION LITERATURE

Moving away from relying solely on proxy measures, a number of studies have stressed the importance of understanding QoL from the perspective of children themselves. Policies at the turn of the century calling for the inclusion of the child in decision-making about their care and Tx (House of Commons Health Committee, 1997; Royal College of Paediatrics and Child Health, 2000; Department of Health, 2003), have also made understanding the child's perspective that much more pressing. However, the nature of the child's participation in the decision-making process has received relatively limited attention (Ranmal et al, 2008). Below, the outstanding issues in the child participation literature are discussed.

A. LACK OF STUDIES EXPLORING CHILD'S PARTICIPATION

The child's voice and participation in healthcare remains an under-researched area due to a number of factors.

Until rather recently children were viewed as passive objects of concern in research (Dixon-woods et al, 2005; Bluebond-Langner & Korbin, 2008; Prout & James, 2015). Coyne and Carter (2018) highlighted the ethical and organisational hurdles researchers face when attempting to conduct research directly with children and the ongoing issue of gatekeeping once ethical approval has been secured.

Doctors have not always supported children's participation in consultations, demonstrating protective attitudes and citing lack of time, appropriate communication skills as well as uncertainty about children's competence (Coyne, 2008). Stivers (2001) presented the child's often presumed limited competence as creating '*interactional complications'* which can create dilemmas for clinicians. Two literature reviews concluded that even amongst those who acknowledged the importance of children's participation in decision-making there was disagreement about the degree to which children should be involved (Coyne, 2008; Davies & Randall, 2015). Both literature reviews also called for further research to explore children's participation in consultations and decision-making (Coyne, 2008; Davies & Randall, 2015). This thesis adds to the scarce evidence in this area.

B. RESEARCH 'ABOUT CHILDREN' STILL FOCUSED ON THE CLINICIAN-PARENT DYAD

Coyne's (2008) review of children's participation in consultations revealed that these studies focused heavily on doctors' and parents' facilitation of child participation, the marginalisation of the child in the consultation, and what children did or did not say. The studies would seem to indicate that children were rarely involved in the consultations and decision-making process.

A systematic literature review of decision-making in adolescents with cancer conducted by Day et al (2016) found little attention to the role of the young people themselves in the

consultation and decision-making and called for further research to explore adolescents' place in the consultation and decision-making process (Day et al, 2016). This thesis heeds this call. Chapter 5 explores the participation of a teenage girl through DA. Using this method of analysis along with CA of real-life triad interactions aids the exploration of the child's participation within the real-life consultation.

C. ABSTRACT GUIDANCE REGARDING RECOMMENDATIONS FOR CHILDREN PARTICIPATING IN DISCUSSIONS AND DECISIONS IN THE CONSULTATION

While policies and clinical guidance from the turn of the century (House of Commons Health Committee, 1997; Royal College of Paediatrics and Child Health, 2000; Department of Health, 2003) advocate involving children in discussions and decision-making regarding their health and well-being, they are without recommendations for how to go about doing so. Little guidance exists on how to go about translating these recommendations into practice (Coyne et al 2014) and what guidance exists is often abstract and non-specific. (Ruhe et al, 2016b; Kelly et al, 2017).

D. METHOD USED TO EXPLORE CHILD'S VOICE

While there is agreement in the literature that QoL is a subjective phenomenon and as such, there is a need to elicit the child's voice, there is a question as to how this is best accomplished. To date, most researchers have relied on interviews conducted at particular points in the illness trajectory. Longitudinal studies where different periods in the illness are explored to understand how participation changes over time are lacking. Kaye et al (2018) have advocated recording of real-life consultations, in contrast, to interview studies, calling it a gold-standard methodology for the investigation of communication and decision-making in paediatric oncology. Observation and recording of actual on the ground consultations eliminate such issues as recall bias, inequality in researcher–child relationship, interpretation of the question and allows for the capture of actual experiences of children in real-time as the discussions and decisions unfold. The

'in vivo' approach avoids problems of inaccurate or distorted recall of what occurred in the consultations (Bluebond-Langner et al, 2017) and presents a nuanced and more objective insight into a phenomenon.

Studies based upon recorded consultations have been conducted to understand parent and child participation in consent consultations but typically use quantitative methods, counting the time speaking, number of questions, etc. The studies focussed solely on the child's spoken words and so missed much of the child's voice. For example, while Miller and Harris (2012) explored children's involvement in discussions on consent for clinical trials using observations of real-time consultations, they calculated word count in the consultation, reporting that the level of patient to physician communication was low, only 3.2% of the consultation.

Olechnowicz et al (2002) similarly, using observational methods, explored the child's involvement in discussions over clinical trials, and additionally the child's diagnosis and Tx options, by calculating the percentage coverage of the child's verbal contributions, focusing in particular on the patient's questions.

In contrast, this study, using an interactionist perspective, looks at the child's involvement through an examination of: 1) interactional sociolinguistics, including the participant's different roles and identities, in the context of how roles might be related to or influenced by language. Tates et al (2002b) state that:

> *'participants display their orientations to institutional roles and identities through their use of linguistic devices…[A] clue about participants' orientations to institutional talk is the use of meta-communicative expressions by which people refer overtly to the way they organize their interaction with the others' (p 110).*

For example, in the context of a triadic analysis, Tates et al's (2002a) 'child-centred behaviours' are utilised. 2) differences in the child's contributions within the different interaction types (e.g. child-clinician alone); 3) the child's non-verbal communication; 4) the child's input, i.e., when the child's speech is initiated or prompted and when verbal in English or when translating to Arabic for her mother; 5) the child's questions and decision-making; and lastly, 5) when a parent or clinician states the child's view's and when the child verbalises their own view. Using DA which uses talk that is naturally found (as in

real life observed consultations) and taking note of context, the analysis looked past the word's literal meaning to explore all the different ways the child's voice emerges in the consultation.

E. NARROW VIEW OF CHILD PARTICIPATION

There is no accepted definition of participation (Quaye et al, 2019). Quaye et al (2019) spoke of active participation as '*a transfer of information and power such that the participant's views influence decision-making'* (p.4526). Similarly, Clemente (2009) situates children's participation '*in the specific circumstances within which children's actions occur'* (p.873). However, in practice, participation can mean many things, from receiving information to making care and Tx choices (Miller and Harris, 2012). Recommendations on children's participation range from advocating full involvement, including leading on decision-making, to having the parents or clinician make decisions on the child's behalf, with very little research to support either of these positions (Hinds et al, 2012).

Clemente (2015) warns of the dangers of reducing participation to the observation of whether or not the child speaks. He argues that focusing on verbal expression only is often at the expense of non-verbal expression. Important contributions and how the conversation is co-constructed may be missed if non-verbal contributions are ignored (Clemente, 2015).

Carnevale (2020) mentions the impact of the administrative/legal context on what constitutes children's participation. He argues that recognising agency solely in people who are '*legally autonomous decision makers who can bear responsibility for the consequences for their decisions'* greatly limits who can be recognised as agents and thus seen as able to participate in discussions about their care. The exploration of the child's participation in Chapter 5 is marked by attention to non-verbal behaviour as well as verbal and by an exploration of when the child offers their verbal/non-verbal responses. In conducting these analyses using real-time consultations, the method aims to capture a more fine-grained and wider understanding of participation than is currently limited in the literature.

F. RESTRICTED APPROACH TO ANALYSING THE INTERACTION

Another issue of concern in the child participation literature lies in the approach taken to analyse the interaction between children, parents, and clinicians. A number of studies find a need for further research examining the role of the child in the consultation as a triadic process (Bluebond-Langner & Langner, 2021; Clemente, 2015; Day et al, 2018; Coyne & Gallagher, 2011) and specifically to explore how parents and clinicians can promote child participation in meaningful ways (Clemente, 2009). Studies to date have mostly focused on and analysed dyads in the consultation only or the child on their own (Tates and Meeuwensen, 2001). In their review of the literature, Tates and Meeuwensen (2001) found that most studies exploring doctor-parent-child communication only focused on dyads (child-clinician or parent-clinician) within the interaction. Their review concluded that triadic analysis is needed to fully understand children's views. Gabe et al (2004) also stressed the importance of including all three perspectives in the analysis of paediatric consultations to enable a '*situationally contextualised view of partnership'*. Weaver et al (2015) and Day et al (2018) also concluded that the child's involvement in the consultation is an interactive process and should be analysed as such.

In line with these recommendations, this study used the methods of DA and CA aligning with an interactionist perspective, looking at all participant groups in the interaction and using verbatim transcripts of actual consultations.

G. RECENT CONTRIBUTIONS TO RESEARCH ON CHILD PARTICIPATION

Research conducted since Day et al's review has found variability in the experiences of children with cancer in consultations. Kelly et al (2017) found children's participation preferences could be summed up by the phrase 'having a say, as I need at this time', indicating children's need for flexibility in their involvement. Children generally wanted more say in the smaller or minor decisions in the consultation and relied upon and trusted the adults to help them in the consultation (Kelly et al, 2017; Coyne et al, 2014).

Ruhe et al (2016a; 2016b) and Weaver et al (2015) also found children with cancer reported varying involvement in health care discussions. Children differed in the amount

they participated in discussions and decisions in the consultation depending on the time, situation, social context, or decision. Weaver et al (2015) referred to children's decisional involvement as on a spectrum ranging from a preference for being fully involved to a preference for the children to be passive.

Zwaanswijk and colleagues (2007) used focus groups to explore the communication preferences of children with cancer, survivors, and their parents. Authors found that all participants preferred a collaborative role, however, children reported that there were times they felt they had no part in a decision due to the availability of a prescribed Tx protocol they felt they had to take.

These studies provide evidence of the complexity of children's preferences concerning involvement in decisions and the consultation generally. 'Where' and how the child wishes to be situated can change across the course of a consultation. They also confirm the issue that was raised in the previous section, that consultations involving children (and likely often with adults) need to be approached as a triadic (or N-adic) interaction. The demonstration, significance and implications of the points raised in these studies are at the core of the close analysis of the case presented in Chapter 5.

2.3 CHAPTER CONCLUSION AND TRANSITION TO CHAPTER 3

This chapter aimed to situate the thesis within the most relevant literature in children's QoL research, that of relevant issues in the QoL literature and relevant issues in the child participation literature. There is support for research exploring the subjective and dynamic phenomenon of QoL as it occurs in real-time and over time. The limited previous studies on child participation stress the need to interrogate the child's voice in the QoL discussions by employing a broad understanding of participation and exploring non-verbal as well as verbal behaviour. Previous research recommends looking at the triadic interaction from all three perspectives involved in the consultation to understand how the participant groups interact. Lastly, from previous research, it was clear that a method was required which not only realised the values of an interactionist approach but also allowed for an open analysis.

CHAPTER 3 – METHODOLOGY

3.0 CHAPTER OVERVIEW

This thesis uses a variety of qualitative methods to explore how QoL figures into clinical consultations for children with an HRBT. This chapter presents the methods used for analysing the transcripts from the HRBT project and the rationale for selecting these approaches.

The chapter begins by stepping back to describe the wider HRBT project which provided the data for this thesis as well as for a number of other investigations. It then presents the theoretical perspective, methodological approach of the thesis and finally the research method and analysis for each of the two empirical chapters: Chapter 4 - Quality of life in children with high-risk brain tumours according to four domains: Parents' and clinicians' perspectives over the illness; Chapter 5 - The child's voice in quality of life discussions as revealed in clinical consultations.

3.1 – HIGH-RISK BRAIN TUMOUR (HRBT) PROJECT

This section describes the HRBT project which provided the data for this thesis. It outlines the setting of the study, the recruitment and demographics of the sample, the ethics approvals for the study, some background on ethnographic research, and finally the methods of data collection.

A. STUDY SETTING OF HRBT PROJECT

Research for the HRBT project was conducted at a joint university hospital and a tertiary paediatric hospital. The paediatric hospital sees approximately 100 (out of 400 in total across the UK) new patients with Central nervous system (CNS) tumours a year, ~20 % of whom have *high-risk* BTs. The joint service provides inpatient and outpatient care for 0-24-year olds diagnosed with BTs and other solid tumours.

B. SAMPLE AND RECRUITMENT OF HRBT PROJECT

The consultations were seen as taking place between three participant groups: the patients, children with HRBTs; their parents and additional family members; and healthcare providers. Recruitment proceeded based on the identification of an eligible child. Their family members and the healthcare professionals (HCP)/clinicians who provided their care constituted the other participant groups but their characteristics were not relevant to recruitment.

Children and their families

All children were selected from the study hospitals as noted above. All paediatric patients discussed at the weekly specialist neuro-oncology multidisciplinary team meeting (MDT) with a diagnosis of high-grade glioma, diffuse intrinsic pontine glioma (DIPG), atypical teratoid rhabdoid tumour (ATRT), or high-risk embryonal tumour (formerly designated as high-risk CNS primitive neuroectodermal tumour) were eligible for the study. During 20 months of data collection, newly diagnosed patients and families were identified by clinicians as eligible for the study at the neuro-oncology MDT. Eligible families were approached sequentially about participation unless ethnographer capacity limited new enrolments. Newly diagnosed cases were approached only if ethnographer resources could accommodate an additional case at the time of diagnosis. Nineteen out of 25 families approached gave written consent to participate in the HRBT project (see Table 1 in Chapter 3 for sample demographics).

Clinicians

In the main, clinicians in the study were from neuro-oncology, radiation oncology and neurosurgery as well as the paediatric palliative care team. The latter are comprised of specialty paediatric palliative care consultants and clinical nurse specialists and are complemented by a large multidisciplinary team and additional ward staff. This thesis focuses on consultations with the oncologists and neurosurgeons in the hospital and consultations with the palliative care team consultants and clinical nurse specialists at the child's home, from entry into the study through death or the end of 20 months of data collection.

Oncologists, radiotherapy and surgical (disease-directed) HCP were grouped together, as were palliative care clinicians. This was a decision made after analysis began and it became clear that there were some differences between the two groups. Where there were differences between disease-directed HCP and PC clinicians, this was highlighted.

TABLE 1. SAMPLE CHARACTERISTICS FROM HRBT PROJECT

Characteristic		**Total number of cases in the sample**
Diagnosis	ATRT[a]	4
	DIPG[b]	3
	High-Grade Glioma	4
	High-risk embryonal tumour[C]	8
[a]Atypical Teratoid Rhabdoid Tumor; [b]Diffuse Intrinsic Pontine Glioma; [C](previously called high-risk CNS primitive neuroectodermal tumour)		
Disease stage at entry into the study	Diagnosis	14
	1st progression	3
	2nd progression	2
Number of children for whom data were collected at each stage of their illness [Note: some participants had fewer progressions prior to death than others].	Diagnosis	14
	1st progression	7
	2nd progression	6
	3rd progression	4
	4th progression	2
	Death	9
Gender	Male	10
	Female	9
Age range	0.9 years – 15.7 years; Median 5.5 years)	
Age at entry into the study	<5	10
	6-10	6
	11-16	3
Parental gender	Mothers	17
	Fathers	15
Language spoken in the home	1 Albanian, 3 Arabic, 1 Bangladeshi, 1 Dinka, 6 English, 1 Mandarin, 1 Polish, 3 Somali, 1 Sylheti, 1 Urdu	
Households speaking only English		6
Households speaking English and another language		13
Religion of the child	Muslim	9
	Unknown	4
	Christian	4
	Buddhist	1
	Catholic	1

C. ETHICS AND INSTITUTIONAL APPROVAL OF HRBT PROJECT

Data collection methods and procedures were reviewed by the Patient and Public Involvement Group for the HRBT project. All methods and procedures were found to be acceptable. Advice and support were sought and received from the UK Health Research Authority Confidential Advisory Group. The HRBT project was approved by the Bloomsbury Research Ethics Committee and the Research and Development departments at both study hospital sites in December 2013.

D. HRBT PROJECT AS ETHNOGRAPHIC RESEARCH

The HRBT project was conducted as an ethnographic project. The choice of method was a consequence of the research aim of the project which was to understand how all participants approach and contribute to decision making, through a close examination and analysis of the actual encounters as they occur throughout the entire course of the child's illness and Tx. Ethnographic research can be viewed as the '*first-hand experience and exploration of a particular social or cultural setting on the basis of (though not exclusively by) participant observation*' (Atkinson et al, 2001, p.4). The project was designed to capture data longitudinally and continuously, a task for which ethnography is well suited.

The encounters on which the project focused were clinical consultations primarily between the family and oncology consultants and also with the palliative care team. These consultations were to be subjected to close analysis which made accuracy essential. Based both on literature and the PI's (Bluebond-Langner) decades of experience attending consultations and subsequently discussing them with participants it was decided that retrospective interviews were not reliable for the purpose of capturing the consultations in the detail required. Two ethnographers undertook the task of attending and recording the consultations while also taking hand written field notes on non-verbal behaviour. They were introduced to the families as researchers and were thus assigned a role in the consultation.

Some informal conversations were recorded and some interviews were conducted with participants, but these were not done systematically and with all participants.

Ethnographic studies have provided a foundation for studies in a range of fields, particularly in healthcare where vulnerable participants and sensitive situations can be followed, as the following examples illustrate. Bluebond-Langner's (1978) pioneering ethnographic work that explored the experiences of dying children in America is an example of the insights that participant observation can provide about otherwise unknown and sensitive phenomena. Bluebond-Langner was able to give a voice to previously unheard children with leukaemia and provide a rich presentation of their journeys. Bluebond-Langner et al (2017) highlight the importance of the naturalistic methodology participant observation offers: *'In taking an 'in-vivo' approach, problems of inaccurate or distorted recall from retrospective interviewing are avoided. The complex interchange is documented as it unfolds'* (p.469).

E. METHODS OF DATA COLLECTION IN HRBT PROJECT

The HRBT project adopted a participant observation methodology that involved the audio recording of the encounters to explore ways in which decision-making, and indirectly QoL, figured into consultations about the care and Tx of children diagnosed with an HRBT. It involved embedding researchers into the clinical consultations, establishing them and immersing them in the clinical context of care and Tx of children and families. This process was facilitated by having clinicians introduce the ethnographers as carrying out clinically relevant research which would apply to children such as those approached for the study. The researchers attended, audio-recorded, and took notes on as many interactions as possible, among parents, patients, and clinicians over 20 months (February 2014 – October 2015). The ethnographers carried their audio-recorders round their necks to provide as much transparency to all participants about their presence as possible. Ethnographers carried notebooks with them at all times and wrote fieldnotes to accompany the audio-recordings and augment the verbatim transcripts. The fieldnotes were non-interpretative, were guided by a template and included who was speaking to whom, non-verbals and where people were seated. Inevitably there were still encounters that were missed. Some missed encounters were found out retrospectively, e.g. a child would have a last-minute appointment or visit booked in and the clinicians forgot to let the research team know, but the research team was informed by the family or clinician after

the event. Or, after the study had been open for many months, when many cases were being followed simultaneously, ethnographers would have to choose to attend and observe one encounter rather than another. There may have also been encounters that the research team never learnt about. The HRBT research team were confident that they covered the overwhelming majority of the clinical encounters experienced by the family and therefore covered most of the discussions and decision-making made by the family and the clinical team.

For some patients, the period in the study covered diagnosis through to death. Interactions included consultations, palliative care team visits at the child's home, hospital and hospice and MDTs. Recordings were uploaded to UCL's data safe haven which could only be accessed by researchers on the project with logins to the safe haven. All recordings were transcribed verbatim in house by one of two transcriptionists employed onto the HRBT project and non-verbal information taken in the fieldnotes by the ethnographer at the time of the encounter was added to each transcript in the form of parenthetical remarks , e.g. '(patient gestures trembling hand)' or '(patient tabs at her lip with a tissue)' by one of the two ethnographers after the verbal transcript was complete. One of the two ethnographers would proof the transcript and sign off as complete and ready for analysis.

Although not one of the main ethnographers on the HRBT project, I was involved in the data collection towards the end of the study period, attending several consultations, MDTs and informal encounters (~six encounters over three different days) and therefore have a concrete insight into the data collection involved in the HRBT project.

The question is sometimes asked if whether participant-observers, by their very presence, affect, and thus alter, the interactions which they observe. There are several reasons why it is believed that this is not a significant concern. The presence of staff other than the treating consultant during consultations is routine. It is also routine for families to encounter unfamiliar HCPs in consultations. Unpublished data from the HRBT project showed that at diagnosis consultations alone, the mean number of medical professionals present during consultations was 3.6 (range from one to seven). The presence of one more person thus became routine. '[I]*n time, both clinicians and families find their presence, wherever it is, unremarkable*' (Bluebond-Langner et al, 2017, p.468). Further,

there is no known reason to expect that the presence of an ethnographer/researcher would alter the consultation systematically, that is in the same sort of way in every case. This is to say that the presence of the ethnographer would not introduce a consistent bias into the interaction.

The sample of the HRBT project, and indeed of most participant observation studies, is small at 19 families. However, as explained earlier in the chapter, ~20 % of the 100 children the paediatric hospital sees a year have *high-risk* BTs and therefore the total of 19 participants in the HRBT project is significant given the total number of children diagnosed with HRBTs in the UK in a year. The small number of participants is offset by the intensity of the study and by a large number of consultations recorded continuously over the duration of the study.

3.2 - THEORETICAL FRAMEWORK – INTERACTIONISM

This section describes the interactionist perspective which provides the theoretical underpinnings for the thesis.

Interactionism is not a specific theory. Atkinson and Housley (2003) argue that interactionism is now a pervasive influence on much social science and qualitative research, and that '*we are all interactionists now'* (p.2).

Some basic principles of interactionist research have been identified. The interactionist perspective regards the social world as a milieu in which meaning is formed during and through interaction between individuals (Rock, 2007). Social reality is an interpreted reality. The interpretations which constitute social reality emerge in interaction, they are joint products and not simply how different individuals might want to regard things. One consequence of this is that individuals using language in interaction are not seen as simply transmitting and receiving information but as active agents in the formation and interpretation of behaviour and action (Bluebond-Langner, 1996, 1978). I maintain this focus throughout the thesis, reflecting on the interaction process between the children, parents and clinicians in principle and practice.

In the interactionist paradigm, ontology and epistemology are linked. In this thesis the position of interactionism is that a shared constructed reality exists for the participant groups (children, parents and clinicians) holding the constructions. Yet these

interpretations, these meanings are not reality in itself, reality is not totally something in the minds of participants in itself but is constructed in response to an independently existing reality which participants encounter only from a situation and not "in itself". Interactions are publicly available and can be observed by ethnographers. Interactionism's position is that individual and shared constructions of reality can be accessed through interaction in naturalistic settings where behaviour can be observed. So, in the first instance, the reality which interactionist research investigates is available only via the participants themselves acting and creating social reality in naturally occurring rather than in staged or controlled situations.

Atkinson and Housley (2003) offer several concepts that help in understanding interactionism, among them is language and communication which is the focus of this thesis. Language, meaning, and communication are at the heart of interactionist work and increasingly, interactionism has featured in the study of discourse (Atkinson & Housley, 2003). Relevant to this thesis and the HRBT project, Rock (1979) claims that '*society is held to emerge from discourse'* and '*interactionism portrays social life as an ongoing series of conversational encounters*'(p.116).

Working within an interactionist perspective, I sought research methods which would be suited to the research question, aims and objectives. Such methods had to be applicable to naturally occurring conversation. Also, since I intended the findings to be of use to clinicians who conduct such consultations it was appropriate to use methods which based analysis upon what was evident within the conversation itself.

Therefore, in this thesis, an approach is adopted that resembles what Antaki (2008) has called generic discourse analysis (DA) (this is not to be confused with critical discourse analysis). The approach is built upon four principles: (1) the talk or text is to be naturally found (as in real life observed consultations); (2) the words should be understood in their context; (3) the analysis should be sensitive to the words' nonliteral meaning; (4) the analysis should reveal the social actions performed (Antaki, 2008). Antaki's third principle, the "*nonliteral meaning*", features heavily throughout the thesis, particularly in the development of the dimensions of QoL (see section 3.6 below). Our analyses aim to satisfy the requirements which Antaki proposes:

> '*Any discourse analyst who claims to be analysing …must 'add value' to what is readable or hearable in the words straight off, beyond simple paraphrasis or glossing; they must be able to back up their claims with some evidence grounded in the words used or warrantably not used; and they must reach their conclusions by argumentative steps available to a fair-minded fellow-scholar*' (p. 444).

In the data analysis, DA principles are in play, talk has been naturally observed, and evidence for analysis is grounded in the language actually used such as in going beyond the words' nonliteral meanings for example in the 'Responds "*I don't know*"' participation code. These principles reflect an interactionist perspective which underpins the thesis. For example, In Chapter 4 an approach is used to develop the dimensions of QoL and document their dynamic appearance in the consultations which is faithful to the 4 principles of DA. In Chapter 5 DA and aspects of conversation analysis (CA) are used to identify and explore the child's voice, be it verbal or non-verbal, in an interaction and from that to amplify the child's views on QoL.

3.3 RESEARCHER POSITIONALITY AND REFLEXIVITY

A way of increasing transparency over how research and analysis is conducted is through disclosure of a researcher's positioning. Savin-Baden & Major (2013) identify three main ways that a researcher can identify their positionality: locating in the subject, locating themselves about the participants and lastly locating themselves about the research context and process. Regarding the subject, I have worked in research with children for 13 years and specifically research involving children with life-limiting illnesses for 11 years. I am a white, British female who only has English as a language spoken at home. I started in this area of research with no children of my own but entered motherhood after a few years. At the beginning of the PhD I had one child of toddler age and during the PhD, had another child. Having children of my own whilst exploring this sensitive and emotive subject area has affected the way I understand and explore these families' lives. It certainly has been harder emotionally and I feel I have more empathy for the families since having children of my own.

Regarding the participants, having only English as a language I will inevitably miss some of the cultural nuances of this highly diverse population and my culture as a British born person will influence my interpretations of these diverse families.

Finally regarding the research context, my background in Psychology and more recently sociology and anthropology will impact the methods of analysis. My background having had health problems myself and being in and out of hospital since I was a teenager will influence the insider-outsider (emic and etic) dialectic in ethnography. My experiences are not comparable with the experiences of the families in this study but my time spent in hospital and on medication and Tx will inevitably influence my interpretation of the data in this PhD. This paired with my small experience of participant observation on this study results in what could be interpreted as a straddling of the insider-outsider position.

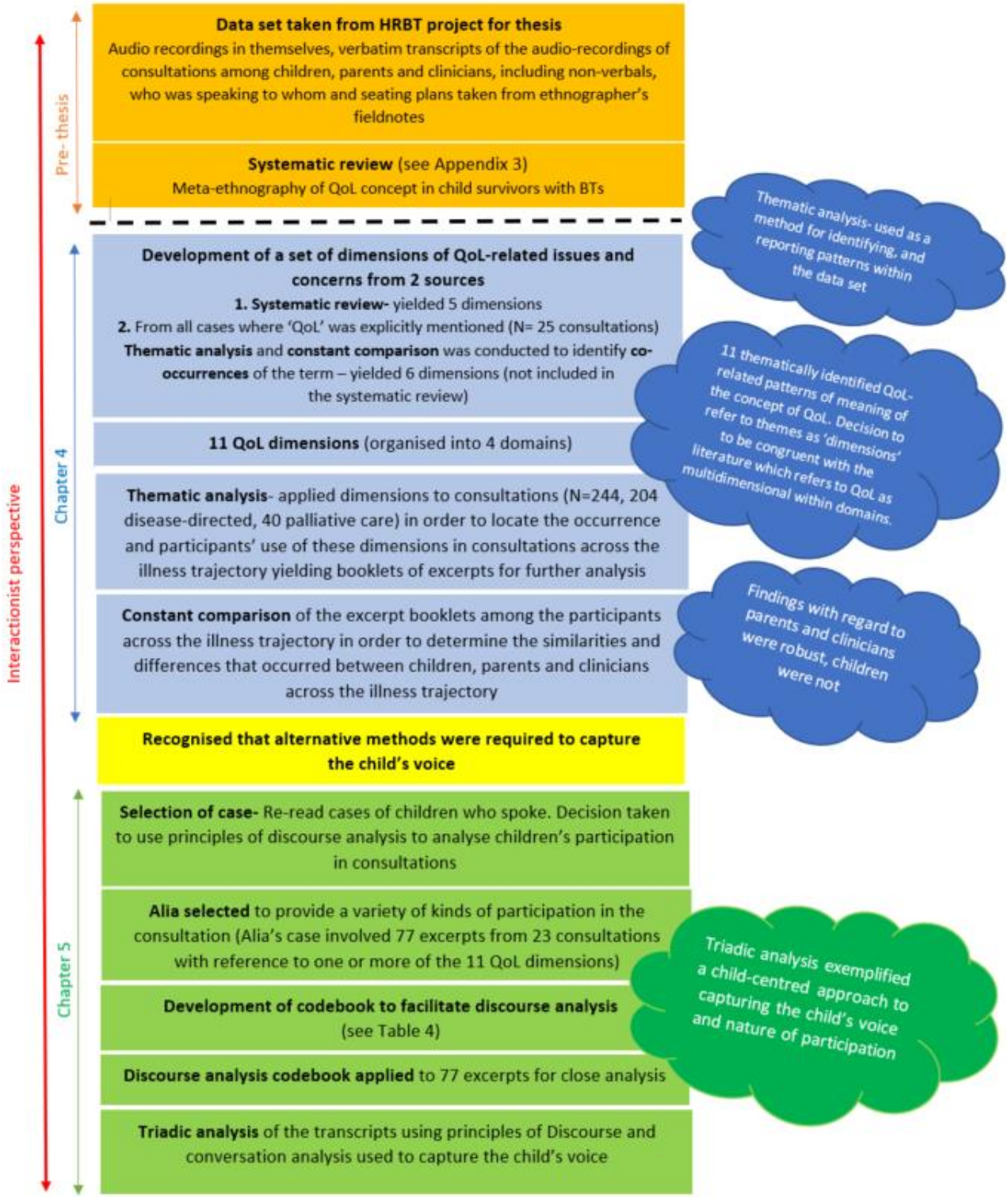
3.4 – ETHICS AND INSTITUTIONAL APPROVALS OF PhD PROJECT

Approvals for my thesis project were gained at the start of the program. The Research Ethics Committee (REC), R&D approval and other institutional approval applications for this PhD project were initiated in December 2015 and were distributed to the relevant committees for approval in February 2016. This included registering the study with data protection, completing risk assessments and insurance documents, and applying for ethical review and approval from UCL ethics and the study hospital's R&D department. In March 2016 the approval process for this study was completed and permissions were granted from the UCL university ethics committee and the university and paediatric hospital's Joint Research & Development Office (reference number: 16PP02).

3.5 – METHODS USED IN THE DEVELOPMENT OF THE THESIS

There were different methods used for each of the results chapters (Chapters 4 and 5). The specific methods for each chapter are explained in sections 3.6 and 3.7. Figure 2. below is an outline of all the methods used in the development of the thesis.

FIGURE 2. METHODS USED IN THE DEVELOPMENT OF THE THESIS



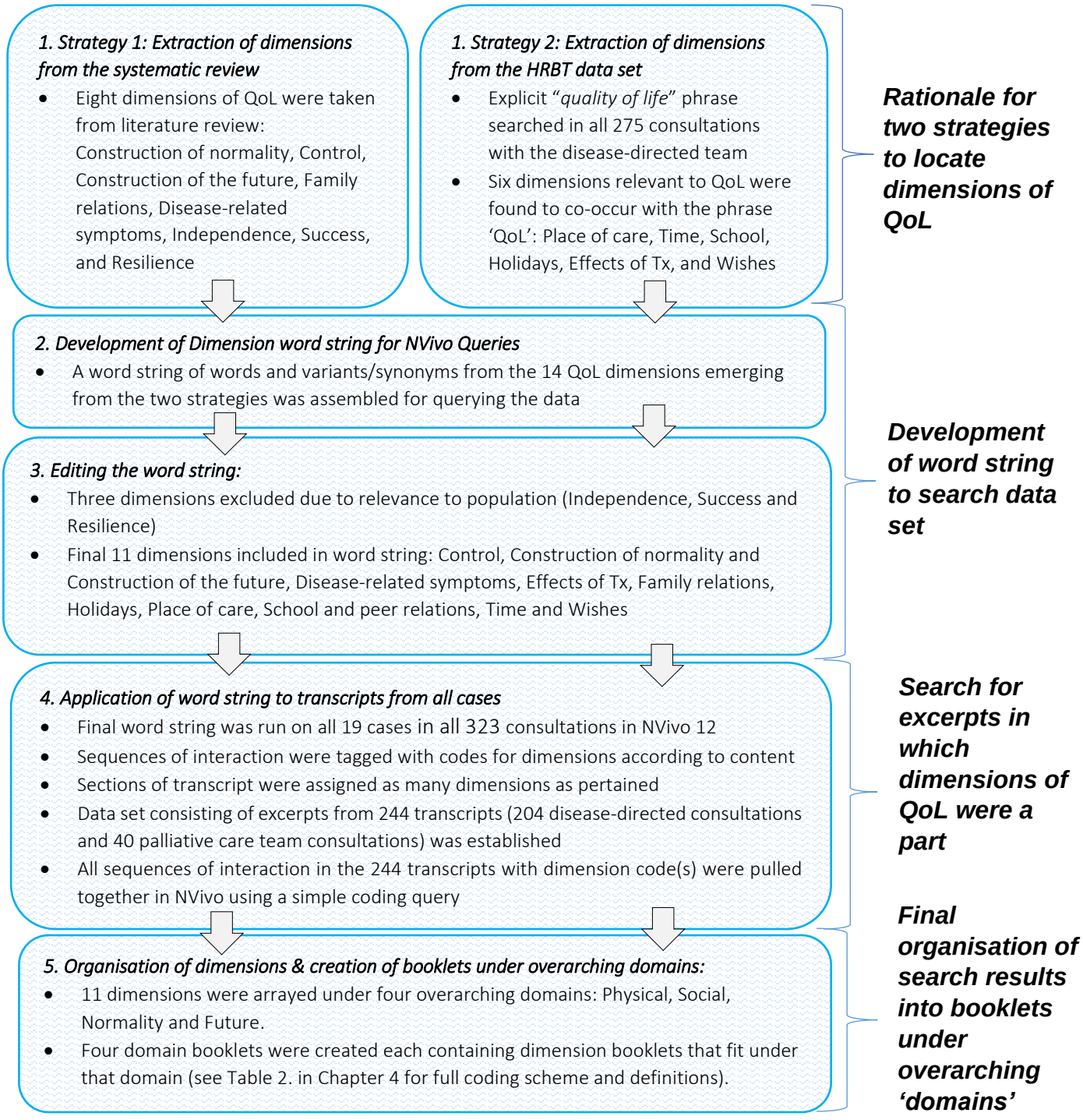
3.6 – METHODS FOR CHAPTER 4

APPROACH AND PROCEDURES FOR THE EXPLORATION OF THE ISSUES AND CONCERNS THAT SURROUND QOL - CHAPTER 4

I now outline the specific methods and analysis employed in Chapter 4, which explores the issues and concerns that surround QoL for all three participant groups.

Transcripts for the HRBT project number in the tens of thousands of pages. Exploration of the issues and concerns surrounding QoL for all patients, parents and clinicians therefore required a systematic yet flexible approach in order to identify when the issues and concerns around QoL were present in the consultation and then to capture those excerpts for further analysis (see Figure 3 in Chapter 3 below).

FIGURE 3. PROCEDURE FOR THE DEVELOPMENT OF THE BOOKLETS OF EXCERPTS OF CONSULTATIONS IN WHICH QOL ISSUES & CONCERNS ARISE



1. Strategies Developed to Support Initial Stages of Exploration

Two strategies were developed to support the initial stages of exploration. One made use of what was learned from the systematic review of QoL in children with BTs (Beecham et al, 2019), the other made use of the results of the search of the transcripts of the consultations in the data set (see Figure 3 in Chapter 3 above).

The systematic review revealed a new conceptual model of QoL for children with BTs, where a new normal was the key dimension. In the end, five of the dimensions identified in the model were deemed relevant to this population and were used in this PhD project as five of the 11 dimensions of QoL: Construction of normality, Construction of the future, Control, Disease-related symptoms and, Family relations (the other six dimensions were Effects of Tx, Holidays, Place of care, School/peer relations, Time and Wishes taken from the second strategy outlined below). Three of the dimensions in the systematic review were not used in this project: Independence, Success, and Resilience. This probably reflected the difference in the samples in the articles in the systematic review and the sample for this project. The systematic review consisted of studies including mainly adolescent survivors of BT, whereas the sample for the HRBT project consisted mainly of young children with poor prognosis HRBTs. Given the difference in populations it was important not just to use dimensions of QoL from the systematic review but also to look within the HRBT project data for other possible dimensions of QoL.[1]

Transcripts from the HRBT project were handled in NVivo within the UCL data safe haven which is a heavily encrypted system used to store patient identifiable data. The HRBT wider project had its own folder and files within NVivo. I had a separate NVivo file with all the transcripts that related to my PhD project. Because of the different parts to the analysis, the transcripts were kept in different clearly labelled files within NVivo e.g. 'Pilot work on explicit QoL phrase', 'QoL 4 domains' and 'Alia participation analysis'.

Using the query function in the qualitative analysis software NVivo 12, the phrase "*quality of life*" and its synonyms from the previously designed and validated Index-codebook for

[1] NB. The 323 consultations from the HRBT project [275 consultations involving the oncology/surgical consultants in the hospital and 48 consultations involving the palliative care team at home] were stored within the qualitative analysis software NVivo 12 QSR International.

the wider HRBT project (see Appendix 5) were text-searched in NVivo in the 275 verbatim transcribed disease-directed consultations from the wider HRBT project. These included consultations with the treating surgical or oncology consultants. Twenty-five occurrences of QoL or its synonyms were retrieved from 16 consultations involving 10 cases from the HRBT data set.

All 16 consultations were closely and carefully read and re-read. For each occurrence of the phrase QoL, the extent of context (of how much of the transcript was 'tagged' or coded in NVivo as 'QoL') was determined on an instance (occurrence) by instance basis but aimed to include the broader context (discussion before and after the QoL dimension was discussed). The 25 excerpts where the term QoL or its synonyms appeared explicitly were then explored in more detail. To do this, the 25 excerpts were coded using the same codebook (see Appendix 5) that was previously developed to understand the themes occurring in the wider HRBT project.

The 25 excerpts were 'attribute' coded and index-coded (I-coded). Attribute coding and I-coding enables queries to be run quickly in the early phases of analysis without drawing premature conclusions to identify when a conversation occurred and who was involved.

An attribute is a characteristic of something or someone. Attributes used in the HRBT project and this thesis were: the case (patient), diagnosis, speaker and period in the illness trajectory. As a starting point the trajectory was divided into five periods based on Bluebond-Langner's previous work in cancer and cystic fibrosis (1978, 1996, 2017). The five phases are: Diagnosis-1st Tx, 1st Tx – 1st recurrence of the disease, after 1st recurrence, last recurrence before death, end of life (EoL).

I-coding is simply a way of open-coding or indexing the data set. They are non-interpretive codes and are used much as one would an index in a book to locate where in the data set particular information/data topics can be found (e.g. 'chemotherapy' and 'survival/length of life') (see Appendix 5 for I-codebook).

EB, RWL and MBL each independently coded 20% of the excerpts. Researchers resolved disagreements in consultation and revised the codebook to remove ambiguity. After 20% of the data set was coded, EB continued to code the whole data set and checked in

regularly via weekly meetings with RWL and MBL with any queries needing to be discussed. Examples and labelling of codes and subsequently dimensions of QoL were debated until a consensus was reached.

Memos were written throughout the process to record ideas, themes emerging, and questions supporting the development of the QoL dimensions. Glaser (1965) recommended writing memos alongside explicit coding, to reflect on the coding and develop ideas for further inquiry and analysis (see Appendix 6 for example memo). Memos are a useful tool in qualitative research and as Birks et al (2008) suggest:

> *'Memoing serves to assist the researcher in making conceptual leaps from raw data to those abstractions that explain research phenomena in the context in which it is examined' (p.68).*

Key themes co-occurring with the explicit term QoL were explored within and between consultations using the constant comparison method, looking at all examples of the themes identified and also at the cohort of 16 consultations as a whole. From the *a priori* codes and the inductive memos, the dimensions to emerge from this stage of analysis were: **Effects of Tx, Holidays**, **Place of care, School/peer relations, Time,** and **Wishes.**

2. Development of word string to search data set

A word string of words and variants/synonyms from the 14 QoL dimensions (eight dimensions from the review, six from the first phase analysis of the search of the explicit term QoL and its variants/synonyms) emerging from the two strategies was assembled for querying the data. For each dimension, variant terms were included in the word string. For example, in the Construction of normality dimension, the terms in the word string were: normal, normally, normality or normalcy.

3. Editing the word string

At this point, time was taken to reflect upon whether all of the terms in the word string were appropriate for the sample accrued for this study. Half of the children in the study were under five years of age, and there were only three adolescents (see Table 1 in Chapter 3 above). The life expectancy at Dx for children recruited was less than two years. With these parameters, it was decided that some proposed dimensions were more applicable to survivors and the decision was taken not to include three dimensions from the word string: Independence, Success and Resilience. Independence is an issue faced by adolescent and young adult survivors as individuals who are dealing with transitioning into adulthood. Resilience is also a trait apparent during longer-term survival, as is Success. This left 11 QoL dimension 'candidate' terms (five from the systematic review by Beecham et al, 2019, and six from the analysis of the Data Set).

4. Application of the word string to identify QoL excerpts

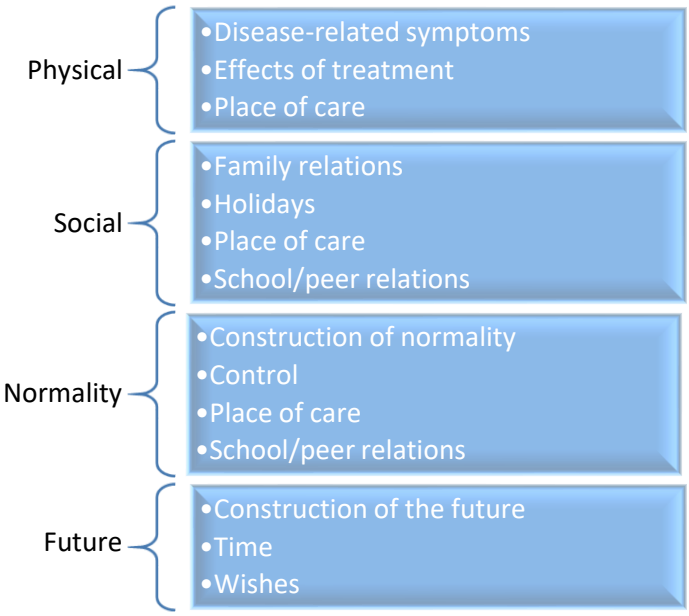
The final word string query including 61 terms and phrases representing the 11 final QoL dimensions in the analysis (see Appendix 7 for NVivo search string) was then run on all 19 cases (323 consultations [275 consultations involving the oncology/surgical consultants in the hospital and 48 consultations involving the palliative care team at home]) which were stored and handled in NVivo. Non-QoL occurrences were identified and removed. EB, RWL and MBL agreed the criteria for the amount of transcript before and after each occurrence of a dimension to include in an excerpt. This process yielded a series of excerpts in NVivo from consultations within each case, with the excerpts each identified by one or more of the dimensions (multiple dimension codes could be applied to the same sections of the transcript as appropriate).

5. ***Organisation of Dimensions under Domains and Creation of Booklets***

After agreeing the 11 dimensions- Construction of normality, Control, Construction of the future, Disease-related symptoms, Effects of Tx, Family relations, Holidays, Place of care

(POC), School/Peer relations, Time and Wishes- dimensions were organised under four domains Physical, Normality, Social and Future (see Figure 4 in Chapter 3 below).

FIGURE 4. QOL DOMAINS AND DIMENSIONS



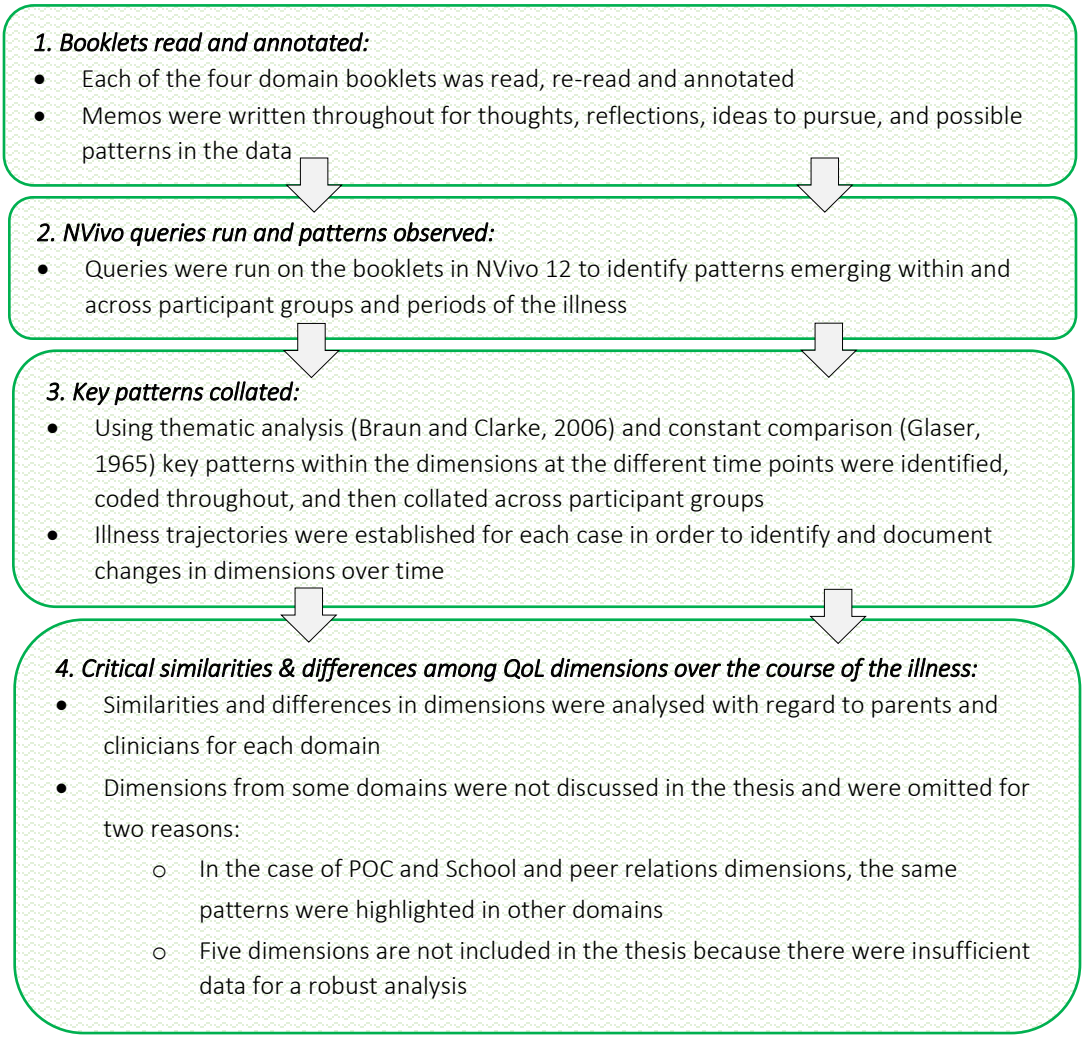
Textual excerpts associated with each QoL dimension were assembled using NVivo, under the appropriate domain. For example, Effects of Tx excerpts were arrayed under the Physical domain, School/peers under Social Domain. Each domain represented a distinct and substantially independent analytic concept, whereas dimensions could fall under more than one domain (see Table 2 in Chapter 4 for definitions of each dimension). At the same time, the fact that individual dimensions could fall under more than one domain indicated that dimensions themselves were multi-faceted with some facets of a single dimension best explored in one category and some in another. For example, the dimension POC appears in more than one domain – Social, Normality and Physical. There are facets of POC that raise issues related to the physical, some to the concept of normality, and others to the social.

Each of the four overarching domain booklets consisted of multiple dimension booklets of excerpts from 244 consultations which became the final dataset for the analysis of

Chapter 4- parent and clinician views (204 consultations involving the disease-directed team and 40 palliative care team consultations at home). The 204 disease-directed consultations took place in the hospital with an oncologist or surgeon and were on the whole shorter in duration than the 40 PC team consultations that took place in the patient's home, with clinical nurse specialists and on the whole involved more family members. The organisation of dimensions into domains was necessary to deal with the enormity of data the thesis was dealing with. Grouping the dimensions under 'domains' also aligned with much of the previous literature as summarised in a systematic review of QoL measures (Eiser and Morse, 2001a).

Each of the four domain booklets was analysed from an interactionist perspective, making use of approaches and tools used in thematic analysis (Braun & Clarke, 2006) and constant comparison (Glaser 1965) with attention to participant group (i.e. patients, parents, clinicians) and by period of the illness (See Figure 5 in Chapter 3 below).

FIGURE 5. PROCEDURE FOR THE ANALYSIS OF THE BOOKLETS OF EXCERPTS ORGANISED UNDER FOUR DOMAINS



Analysis was an iterative process. First, the booklets were read and annotated for key patterns identified for each dimension within each domain with excerpts attribute coded in NVivo for case (patient), diagnosis, speaker, and period of the illness. This allowed for any excerpt pertaining to an issue or concern regarding QoL to be located and linked by attributes and retrieved for constant comparison and recognition of possible patterns. Memos were written throughout the process for thoughts, reflections, ideas to pursue, and possible patterns in the data.

2. NVivo queries run and patterns observed

'Matrix queries' (a query which enables the exploration of coding intersections between multiple codes) were run on the booklets in NVivo to identify patterns emerging within and across participant groups and periods of the illness. These involved the different dimensions run across the different time periods and across different participant groups. This gave an a priori structure to the analysis as dimensions were then explored by time period.

1. Key patterns collated

By exploring results of these queries, using thematic analysis (Braun and Clarke, 2006) and constant comparison (Glaser, 1965), patterns were identified within the dimensions across the time periods. For example, looking again at the dimension 'Construction of normality', the reduced excerpts of the dimension were explored thematically on the data that constituted the reference to normality in the excerpt. Early memos suggested that these different types of normality could be divided into three groups of normality; Construction of normality before diagnosis, Construction of normality post-diagnosis, and no longer normal.

The excerpts were then re-read with these patterns in mind; and the view's held by children, parents, and clinicians regarding these three groups of patterns were compared and contrasted, coded across participant groups and in all excerpts and then documented.

Again EB, RWL and MBL independently coded 20% of the excerpts to ensure the reliability of the groups within the dimensions. Some of the dimensions, rather than differing in nature and having different patterns within a dimension as in the Construction of normality dimension, became more or less prominent as a whole in the participants' QoL concerns, e.g. Effects of Tx (see Chapter 4, Figure 7).

From the results issuing from retrieval of excerpts by attribute of period in the illness trajectory, it seemed prudent to develop an illness trajectory for each of the cases and

conduct constant comparison from this vantage point as well. This not only supported the analysis and development of patterns and theory but also provided a more nuanced view of the incidence of the QoL dimensions. The changes documented in the illness trajectories demonstrated that the QoL dimensions weren't a static phenomenon but changed between participant groups and over time. The illness trajectories were developed collectively as a group, and meetings were held regularly to discuss the identified patterns or prominence of the dimensions to reach a consensus on how to break down and/or present the dimensions.

2. Critical similarities & differences among QoL dimensions over the course of the illness

From the illness trajectories, memos and dimensions that emerged for each participant group, similarities and differences in the dimensions were developed using constant comparison for parents and clinicians for the domains. The dimensions discussed in Chapter 4 are Effects of Tx and POC in the Physical domain, School and peer relations and Family relations in the Social domain, Construction of normality in the Normality domain, and Construction of the Future in the Future domain. For three dimensions-POC in the Social and Normality domains and School and peer relations in the Social domain-discussion is omitted and results are summarised in tables and figures in order to avoid repetitive discussions. In the case of the POC dimension in the Social and Normality domains, for example, the same patterns were found as in the Physical domain in Chapter 4 and discussion of the findings in the POC dimension in the other domains would not have developed the argument further.

Five dimensions are not presented in the thesis for the reason that sufficient data was not available for a cogent and convincing analysis. There was not enough data on the children's views on the dimensions to present the children's views, which were almost exclusively represented by proxy reports. This was due in part to the children's ages. Ten of the 19 were under five years old and therefore did not speak much at all. This contributed to the change in methodology to explore the child's voice, to explore non-

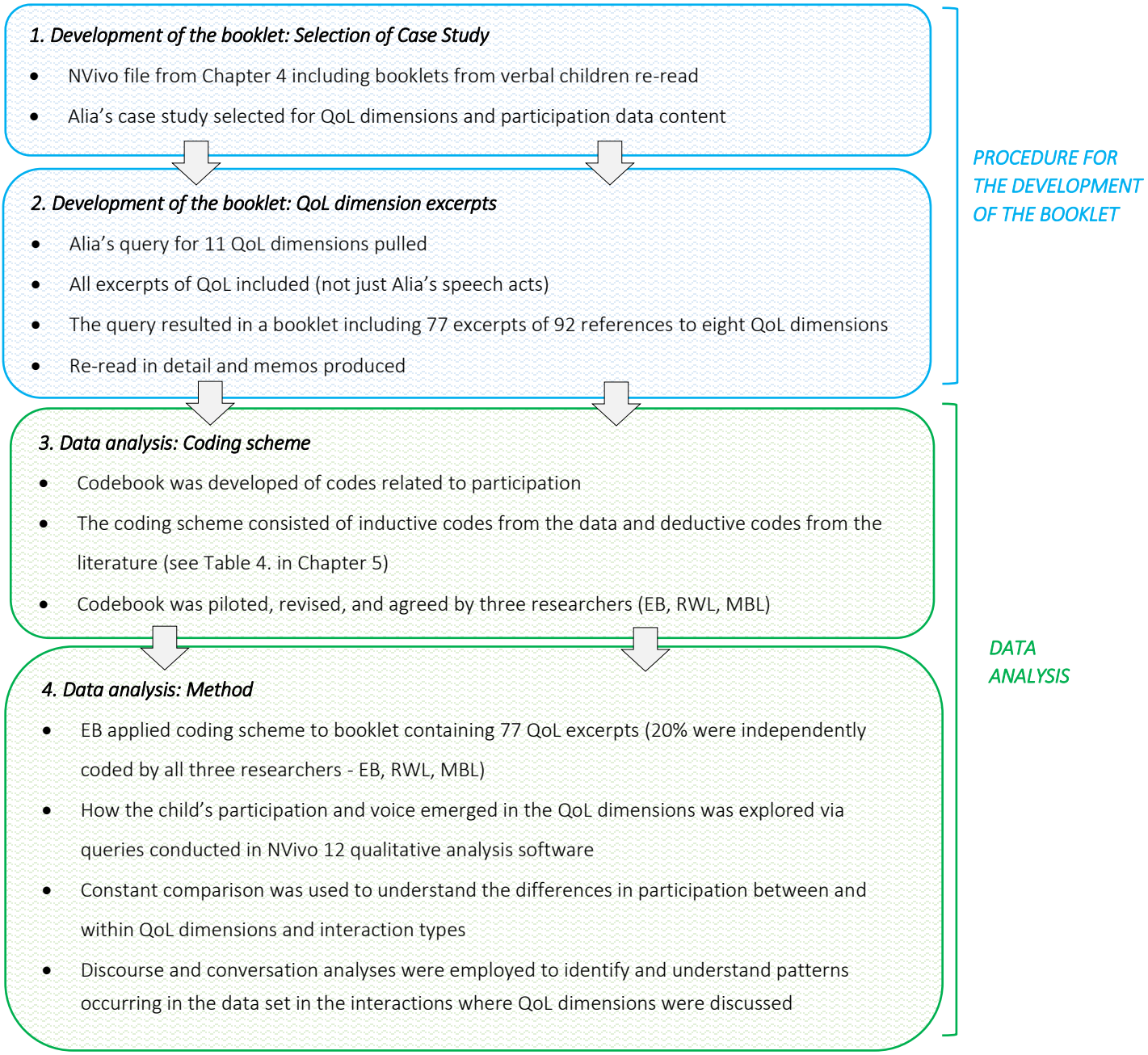
verbal communication, the interaction as a triad, looking at all participants' contribution in the consultation using a teenager who was more verbal.

3.7 METHODS FOR CHAPTER 5

APPROACH AND PROCEDURES FOR THE EXPLORATION OF THE CHILD'S VOICE - CHAPTER 5

Moving the focus of analysis to an exploration of the child's voice, and more specifically their participation in discussions surrounding QoL, presented a number of additional challenges, not least of which was the availability of excerpts in which children participated. Of the 19 cases and 1,263 excerpts from 244 transcripts, 11 children were preverbal or non-verbal, leaving only 8 cases where there was a possibility of excerpts where a child's voice would be accessible. The ages of the 8 children were: 15, 15, 14, 10, 8, 7, 6 and 4 years old. In the first instance, these excerpts were approached in the same way as the excerpts in Chapter 4 (see Figure 6 in Chapter 3 below).

FIGURE 6. PROCEDURE FOR THE DEVELOPMENT OF THE BOOKLET AND DATA ANALYSIS FOR THE CHILD'S VOICE EXPLORATION



1. Development of the booklet: Selection of Case for Analysis

After a re-read of all 8 verbal children's booklets a case was selected because of the amount of participation (verbal and non-verbal) and the variety of participation roles in the interaction. Knowing that presence or absence of a child's verbal participation is not

necessarily indicative of participation (see Chapter 2), a decision was taken to use a case where a variety of forms of participation were available for observation and analysis. The case selected was that of a 14-year-old girl who participated in a number of ways, thereby serving as a means of identifying the range of ways in which children could participate. During 23 consultations over a span of approximately 17 months, Alia spoke for herself and served as a translator for a parent in seven consultations with three different clinicians, often in the presence of siblings and a professional translator as well.

2. Development of the booklet: QoL dimension excerpts

Alia's query for 11 QoL dimensions was pulled and separated from the rest of the cases. All excerpts of QoL were included (not just Alia's speech acts). The data on Alia was 89 pages of 77 excerpts of QoL dimensions extracted from 23 consultations with the disease-directed team. Alia's 77 excerpts included eight different dimensions of QoL. The excerpts were then re-read and memos were produced to explore Alia's participation in the consultations.

3. Data analysis: Coding scheme

A codebook was developed inductively from the data and deductively from the literature to understand how the child participated in the consultation (see Table 4 in Chapter 5). This was piloted, revised, agreed, and then used by the researchers on the QoL excerpts and resulted in smaller participation examples within the QoL dimensions.

4. Data analysis: Method

EB applied the coding scheme to the booklet containing the 77 QoL excerpts (20% were independently coded by all three researchers - EB, RWL, MBL). How the child's participation and voice emerged in the QoL dimensions was explored via queries conducted in NVivo. Constant comparison was used to understand the differences in participation between and within QoL dimensions and interaction types. Features of

discourse and conversation analyses were employed to identify and understand patterns occurring in the data set in the interactions where QoL dimensions were discussed. The findings on Alia's participation in discussions involving QoL issues were presented in Chapter 5 by type of participation.

3.8 – CHAPTER CONCLUSION AND TRANSITION TO CHAPTER 4

This chapter has discussed the theoretical perspective of interactionism and how it relates closely to the methods chosen throughout the study and analysis. The HRBT project that provided the ethnographic data for this thesis was described, and the relational context of this type of data which aligns with the interactionist perspective and the systematic methods to create the dimensions and domains of QoL were outlined. These dimensions of QoL were then used in a wider search to locate all discussions of a broader concept of QoL in all consultations in the hospital and the home. Once the QoL excerpts were found and booklets created for each of the four QoL domains, constant comparison and thematic analysis were conducted on the booklets. The analysis of these natural real-time conversations aimed to reveal the parents' and clinicians' perspectives of QoL as revealed by the dimensions developed (see the next chapter, Chapter 4). Since the data from children was limited, the findings in Chapter 4 were built upon and Chapter 5 was examined beyond the verbal contributions of the child with a wider and more nuanced lens. The exploration of the child's voice was explored in Chapter 5 using a single case, and the QoL excerpts for this one child from the booklets developed in Chapter 4 were explained. A coding scheme developed through a review of the literature and initial analysis of the consultations was then used to explore the child's participation. Methods closely aligned with interactionism were used to analyse the child's participation data. Constant comparison, conversation and discourse analysis were used to interrogate the child's participation in discussions that feature QoL.

In the following chapter, the findings of the analysis of the QoL domain booklets and the key similarities and differences in the QoL concepts between parents and clinicians are presented.

CHAPTER 4 – QUALITY OF LIFE IN CHILDREN WITH HIGH-RISK BRAIN TUMOURS ACCORDING TO FOUR DOMAINS: PARENTS' AND CLINICIANS' PERSPECTIVES, OVER THE COURSE OF THE ILLNESS

4.0 CHAPTER OVERVIEW

This chapter explores how QoL discussion enters into clinical consultations in which decisions about the care and Tx of children with HRBTs are discussed and taken. In addressing this, several assumptions are made; the chapter proceeds on the assumption that QoL is a multidimensional concept. Accepting the concept that QoL is multidimensional, at any given point in a conversation only one or some of the dimensions of QoL may be in play rather than the entirety of the concept. It is also assumed that those participants in a clinical consultation can invoke or speak to QoL issues without explicitly using the terms "quality of life", "well-being", or the like. Also, participants may invoke or rely upon one or more of the dimensions of QoL at different points in clinical consultations.

Analysis will reveal:

(1) The concept of QoL entering into discussions between children, parents, and clinicians through the appearance of specific dimensions of QoL.

(2) How the prominent QoL dimensions change for the parents and clinicians, over the course of the illness.

The chapter is divided into four sections:

Section 4.1 explains the background and methods of the domains and dimensions of QoL used to analyse the consultations.

Section 4.2 examines the critical similarities and differences in perspectives among parents and clinicians, and at different stages of the illness, with respect to QoL dimensions.

Section 4.3 presents the Discussion of findings.

Section 4.4 presents conclusions from the chapter and provides a transition to Chapter 5.

4.1 BACKGROUND/METHODS OVERVIEW

The methods and data analysis for Chapter 4 are presented fully in the previous methods chapter: see Figure 3. in Chapter 3 for an outline of the procedure for the development of the booklets and Figure 5. in Chapter 3 for an outline of the analysis methods.

The way that QoL is dealt with in consultations is explored by identifying occasions where QoL was discussed. With the assumptions described above in view, the first task was to decide upon the dimensions of QoL which would be used in the analysis of the consultations. The next task in the analysis was to locate and extract segments from a large data set of 275 consultations with the oncologists and neurosurgeons in the hospital and 48 consultations with the palliative care team consultants and clinical nurse specialists at the patient's home[2] in which QoL was in play, whether or not it was explicitly named as such.

The dimensions were developed via two strategies; during the systematic review and analysis from consultation data, a search string of QoL terms or 'dimensions' was used to then search for occasions where QoL was discussed throughout the consultations. The dimensions were then organised into a scheme of more specific dimensions and more general domains, with some dimensions taken as specifications of broader domains (see Table 2. in Chapter 4 below). Four overarching domains of QoL were identified: Physical, Social, Normality, and Future, and four corresponding booklets were produced and thematic analysis and constant comparison analysis were conducted using these booklets (see Chapter 3, 3.6). These domains provide the overall organisation for the presentation of the findings in this chapter.

The key or major insights and differences from each domain for parents and clinicians only were extracted and are presented in 4.2 *'Critical similarities and differences of key QoL dimensions over the course of the illness'*. Effects of Tx and POC are presented from the Physical domain, Family relations and School and peer relations were presented from the Social domain, and a Construction of normality and a Construction of the future were presented from the Normality and the Future domains respectively. Due to limited data

[2] Beecham, 'Quality of life' in children with high-risk brain tumours: children's, parents' and healthcare professionals' perspectives over the course of the illness. PhD Upgrade Report, UCL, 2017.

on children and adolescents, Chapter 4 focuses on the major similarities and differences between the parents and the clinicians. Each section in 4.2 starts with an introduction to the domain. The main body of each section outlines how the dimensions change over time for both clinicians and parents, and the sections end with a figure presenting an overview of each domain's major finding/s.

TABLE 2. CODING SCHEME FOR QOL DOMAINS AND DIMENSIONS

Domains	Dimensions	Type/Source of dimension	Description of dimension
Physical	Effects of Tx	Inductive	Any utterance with explicit mention of 'physical', 'side effects' or 'late effects' in relation to the treatment or procedures the child has received or undergone or is yet to receive.
	Disease-related symptoms	A priori/Literature: Beecham et al (2019)	Any utterance with explicit mention of 'symptoms' related to the child's disease including but not limited to: 'seizure' 'somnolence' 'swallow' and 'speech'.
	Place of care	Inductive	Any utterance with explicit mention of 'place of care', 'home', 'hospital' or 'hospice' as a location that the child is or will be cared in by family or healthcare professionals.
Social	School/peer relations	Inductive	Any utterance with explicit mention of 'school' including but not limited to: 'friend' 'peer' 'relationship' 'homework' 'studies' 'nursery' or 'playscheme' in context of friends, not the family and related to the child.
	Family relations	A priori/Literature: Beecham et al (2019)	Any utterance with explicit mention of 'family' including but not limited to: 'sibling' 'brother' 'sister' 'father' 'mother' related to the child's relations with members of their immediate or extended family.
	Place of care	Inductive	Any utterance with explicit mention of 'place of care', 'home', 'hospital' or 'hospice' in relation to the locations as aiding or inhibiting socialising for the child.
	Holidays	Inductive	Any utterance with explicit mention of 'holiday', including but not limited to: 'trip' 'vacation' 'break' 'journey' 'travel' in relation to an extended period of recreation or socialising for the child.
Normality	School/peer relations	Inductive	Any utterance with explicit mention of 'school' including but not limited to: 'friend' 'peer' 'relationship' 'homework' 'studies' 'nursery' or 'playscheme' in context of friends not the family and related to the child.
	Place of care	Inductive	Any utterance with explicit mention of 'place of care', 'home', 'hospital' or 'hospice' as a location that helps or hinders their process of normalisation in the child's life.
	Construction of normality	A priori/Literature: Beecham et al (2019), Bluebond-Langner (1996)	Any utterance with explicit mention of 'normal', 'normality,' 'normalcy,' 'everyday life' or 'usual life' in the context of the child's life.
	Control	A priori/Literature: Beecham et al (2019)	Any utterance with explicit mention of 'control' in the context of influence, management or power of or over the child's life.
Future	Construction of the future	A priori/Literature: Beecham et al (2019), Bluebond-Langner (1996)	Any utterance with explicit mention of 'future' in the context of the child's life.
	Time	Inductive	Any utterance with explicit mention of 'time' in the context of the child's life, including but not limited to: 'time left' for the child, 'precious time' or 'buying time'.
	Wishes	Inductive	Any utterance with explicit mention of 'wishes' in the context of experiences planned by the PC team.

4.2 CRITICAL DIFFERENCES OF KEY QOL DIMENSIONS OVER THE COURSE OF THE ILLNESS

A. PHYSICAL DOMAIN

Introduction to domain

The Physical domain consists of three dimensions of QoL: Effects of Tx, Disease-related symptoms, and Place of Care (POC). Effects of Tx and POC are presented in this section. Effects of Tx relates to the difficulties children experience due to the side effects of the care and Tx associated with the child's disease, and how this talk of the physical effects of Tx features in discussions about the care and Tx of the child. Effects of Tx also encompasses potential late effects of disease-directed Tx. POC as related to the Physical domain involves the Physical location of care of the child; the home, hospital or hospice, and how the location enters into discussions about the care and Tx of the child. Disease-related symptoms relates to the difficulties children experience due to the symptoms of the child's disease itself, but there was insufficient data on this dimension to enable comparison across the participant groups, hence it is not discussed. See Figures 7. and 8. in Chapter 4 below for an overview of the changing perspectives of Effects of Tx and POC within the Physical domain.

At Dx-first Tx

At Dx, oncologists presented the side effects and late effects (Effects of the Tx) of the intended disease-directed Tx as unavoidable and part of the course of Tx (blue quote in Figure 7, below), while at the same time clinicians acknowledged the burden:

> *Dr Toby Mansell: The one other side effect of cisplatin, very important, it can make you feel really sick…we can control all that but we have to tell you what you're up against.*
>
> *Karen Brooke: Okay.*
>
> *Dr Toby Mansell: But again, unless it's really bad, I won't stop any of these medications. The side effects are side effects, you will recover…*

At this early stage side effects are presented as concomitant to Tx. The implicit justification for accepting them is that they are transient and controllable.

> *Dr Louisa Hagan: The idea is to give her the treatment and to try to diminish the side effects of the treatment.*

At Dx, the Effects of Tx featured in the parents' discussions as they began to understand this new world of illness and wished to be prepared as they embarked on caring for their child at home. As one mother remarked:

> *Asalah Kashem: Can I just read the side effects part again?*

Some parents also clearly found the recitation of the side effects difficult to handle, as demonstrated by parents breaking down, crying, or leaving the room. From a parent's point of view, witnessing one's child suffering these side effects is deeply distressing.

POC appeared to be important to some parents at the time of Dx. However, it was important to some parents more than to others. This varied response within the participant group is represented by *two* green lines in Figure 8. The top green line in the prominent cell represents the parents for whom POC was a QoL issue; for example, some parents were reassured by the hospital setting and were happy for their child to be based in the hospital, as illustrated in the discussion with a PC Clinical Nurse Specialist below:

> *Harold Wiseman: So, for this we, we'll be staying here until the 1st, we won't be out between… now and then, will we?*
>
> *Shirley Turner: It depends how she is. I mean some children do go home in between so really, we've got to take it by - day by day.*
>
> *Harold Wiseman: I mean you don't - you're not going to - what I mean is you're not, you're not looking to get us out - do you know what I mean, not, not push us out but I mean –*
>
> *Shirley Turner: No, we won't push you out.*
>
> *Harold Wiseman: - you're not in any rush for us to get out.*
>
> *Shirley Turner: No.*

The lower green line in the 'not as prominent' cell represents those parents for whom POC didn't seem to enter into their QoL concerns:

> *Gurey Dahir: If there's a chance…it doesn't really matter if she's in hospital or not*.

Oncologists emphasised the importance of the child being physically in the home as much as possible throughout the Tx, although oncologists also made clear that during Tx there was sometimes limited flexibility:

> *Dr Louisa Hagan: …He would be at home as much as possible. He will only come in for chemotherapy when he needs to for a day or two and then in the meantime he would be at home. So, we would try to keep him in the hospital the least possible so not interrupt his progress and not interrupt his schooling*…

First Tx-first recurrence

For most parents, concerns about side effects, demonstrated by questioning and airing worries in encounters with HCPs, appeared to decrease after Tx started, as illustrated by the green dashed line moving into the not as important cell in Figure 7. Parents also started speaking about side effects of the Tx as something their child must endure:

> *Dr Toby Mansell: …any other questions?*
>
> *Claire Brooke: No, I don't- I'm just going (to) read up obviously and I just don't need any questions really, just expecting some yucky side effects*.

Two words in this quote, "expecting" and "yucky", point to the change in parental attitude to the Effects of Tx. Side effects have now been incorporated into their everyday life as the parent of a seriously ill child and while unpleasant they are now just one among a number of unpleasant events which one might encounter in daily life.

After the children started Tx, most children were, at various points, required to be an inpatient in the hospital for surveillance. The Tx process itself dictated POC at such times. The fact that remaining in hospital was discussed suggests that POC was still a consideration for the oncologists, although still not prominent at this pre-progression point of the disease. As illustrated in Figure 8, one oncologist informed the patient:

Dr Toby Mansell: Be prepared to-you're not going home. From here (neurology ward) either you have a bit of surgery and then you come to me (oncology ward) or you come (straight) to me. So, you're not going home.

Here the clinician acknowledges that going home after a procedure is likely the default expectation for the parent. At the same time, the clinician indicates that for them it is simply part of the process of Tx.

For some families, PC clinicians entered the support system from this stage in the illness trajectory. Similarly to oncologists, POC didn't drive the discussion or feature prominently among issues raised by PC clinicians at this early stage in the illness.

Even before Tx had started, some parents displayed an eagerness for their child to be at home as much as possible. With Tx underway parents asked for their child to be at home or to come home or delay going into hospital. However, some parents were reassured by the hospital setting and were happy for their child to be based in the hospital. For some parents Tx was their primary concern and they did not express any conflict about being away from home.

After first recurrence

At progression of the disease, the use of dimensions in the Physical domain diverged between the HCPs and the parents (represented by the blue lines that become dashed lines in Figures 7. and 8.). The Physical domain's dimensions, Effects of Tx and POC, appeared as concerns raised by all HCP's (both oncologists and PC clinicians). At recurrence of the disease, oncologists steered parents towards Tx options that had fewer and milder side effects or an experimental Tx with fewer side effects that could be administered at home rather than high dose chemotherapy that had to be administered in the hospital. This can be seen in the quote in Figure 7:

Dr Bruce Simmons: I'd be troubled that giving any anticancer medicine in that circumstance has really got no chance of working and could cause side effects which might make his condition worse…

This reflects a change in the assessment of the burden of Tx.

Parents came into recurrence having heard previously that side effects and late effects were something HCPs wanted to limit, but, implicitly, something that is accepted. At the point of recurrence, and as the child's condition deteriorated, Effects of Tx were not as prominent in the parents' QoL concerns and many parents made minimal mention of side effects. They also reported the child's Physical status as stable. This led to, in some consultations, oncologists and parents debating the child's condition. For example, in most cases, clinicians saw the child as deteriorating, whereas the parent presented their child as a 'healthy sick kid' who should be treated as such (see Normality section).

After recurrence, POC came further to the forefront of the HCP's discussions as a QoL concern (represented in Figure 8. by the blue line becoming dashed and moving into the prominent cell):

> *Dr Bruce Simmons: The side effects with that type of chemotherapy are similar. It doesn't need to be as an inpatient actually.*
>
> *Hodan Omar: uh mmm, yeah*
>
> *Dr Bruce Simmons: One of the benefits of that type of chemotherapy is you can receive the chemotherapy more easily as an outpatient…*

For some parents, the Physical place the child was cared for remained an important dimension of QoL. Yet for others, the location did not seem to be a factor they considered in decision-making. The pursuit of further Tx was the priority and, as before recurrence, if the Tx required hospitalisation this did not deter the parents, implying that POC was not the most important priority for them. The continuation of varied parental viewpoints on POC is represented in Figure 8. by the presence of 2 lines for parents.

Last recurrence before death

At the point of last recurrence before death, symptoms of the disease became increasingly difficult to control and therefore impacted more on the child's day to day life. HCPs often encouraged limiting Tx that caused side effects and still focused on the child being at home.

There were a few parents who, even after multiple recurrences and at the last recurrence

before death, were still relativizing the importance of side Effects of Tx, as the quote in Figure 7. in green implies:

> *Theresa Kiln: Yeah, I think that's what she's got, a resistance to it, I don't think she's having any side effects from it, I (stresses next word) really don't, honestly, hand on heart.*

Most parents, however, were not willing to tolerate the severe side effects at this point when the child had experienced multiple recurrences of the disease and their condition was so unstable (represented by the green line becoming dashed again in Figure 7. and moving back into the important cell).

The somnolence and prospects of having a child in a vegetative state and of losing their social self, as side effects or symptoms, were not acceptable for most parents who had previously accepted the burden of symptoms. Talk about the Effects of Tx and POC increased or re-emerged for these parents in conversations. This discussion of difficult late effects affecting further Tx decisions was observed in one family who discussed possible radiotherapy for their 2-year-old at the last recurrence before death:

> *Anthony Kiln: - yeah, I'm - I, I know how - I think it could be just as devastating seeing her in almost a vegetative state all her life would -*
>
> *Dr Bruce Simmons: Yeah, and, and of course the, you know, it's so difficult when you, you know, you're facing losing a child to, to almost say that anything is better than losing them.*

POC, too, appeared more in conversations for most parents, indicating its importance. QoL was an issue for all parents, indicated by the appearance of QoL dimensions, as the child's condition deteriorated further and as they entered the EoL stage (represented by both green lines becoming dashed and moving into the important cell in Figure 8.).

The emphasis of the oncologists on POC at the last recurrence before death focused on how to keep the child out of hospital:

> Dr Bruce Simmons*: We need to think about where we care for Usman… Obviously he's in hospital at the moment, but if things don't improve it may be that we want to – you would like to take him home, but we provide you with –*

Edith Hutchings*: Yeah.*

Dr Bruce Simmons*: - some support for him at home. But obviously looking at – looking after Usman at home, if he's like this, will be different than it was even a week ago*.

EoL

By the latter stage of the last recurrence, parents and clinicians had re-aligned with respect to both dimensions within the Physical domain (see Figures 7. & 8. in Chapter 4 below).

At the EoL the emphasis of the oncologists on POC continued to focus on how to keep the child out of the hospital, whereas the PC team focused more on the choice of POC. But, regardless of the HCP's preference for a particular POC, they all regarded POC as an important choice to be made at this late stage. Most parents, too, by the EoL, had strong opinions on POC. Most parents preferred to keep their child at home, equating it with what was typical for their family (see Figure 8. below):

Dr Bruce Simmons: The, the other thing that I know that the team would be very happy to do is if, if there's someone that you would want to talk to. Not, not the doctors, not the nurses but someone – 'cause this is really tough. We've got a really good team, psychology team - and I know they'd be happy but - to engage if there's something that you wanted to explore.

Rebecca Jones: (Is crying)

Dr Bruce Simmons: Do you think it might be helpful?

Rebecca Jones: It helps if we go home and be normal.

Dr Bruce Simmons: Can I say though, if you change your mind that they're there because it can be - it, it, and it is literally just someone to talk over when it feels as if - help you cope.

However, some parents, even after multiple progressions, still opted to be based in the hospital for the administration of a new medication. Often their preference, at this point,

was based on practical concerns such as the care of other siblings or inappropriate housing.

FIGURE 7. THE CHANGING PROMINENCE OF THE EFFECTS OF TX FOR ONCOLOGISTS & PALLIATIVE CARE (PC) CLINICIANS AND PARENTS OF CHILDREN WITH A HIGH-RISK BRAIN TUMOUR THROUGHOUT THE ILLNESS TRAJECTORY

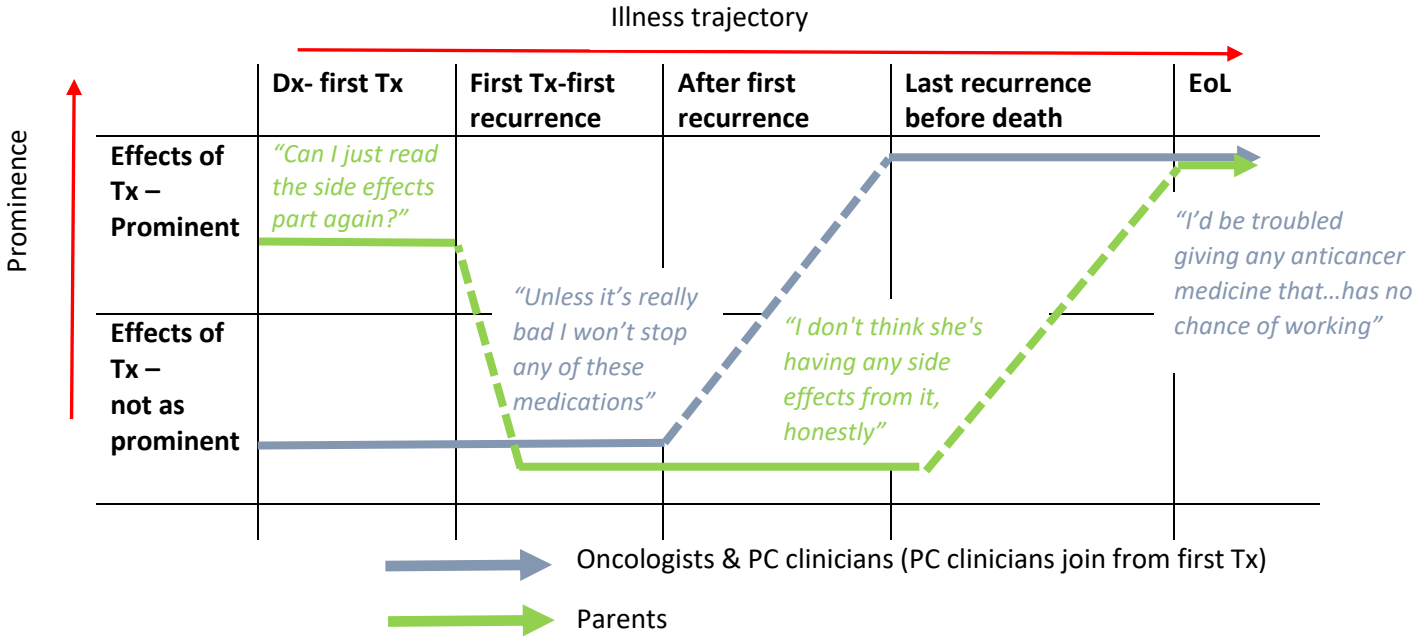
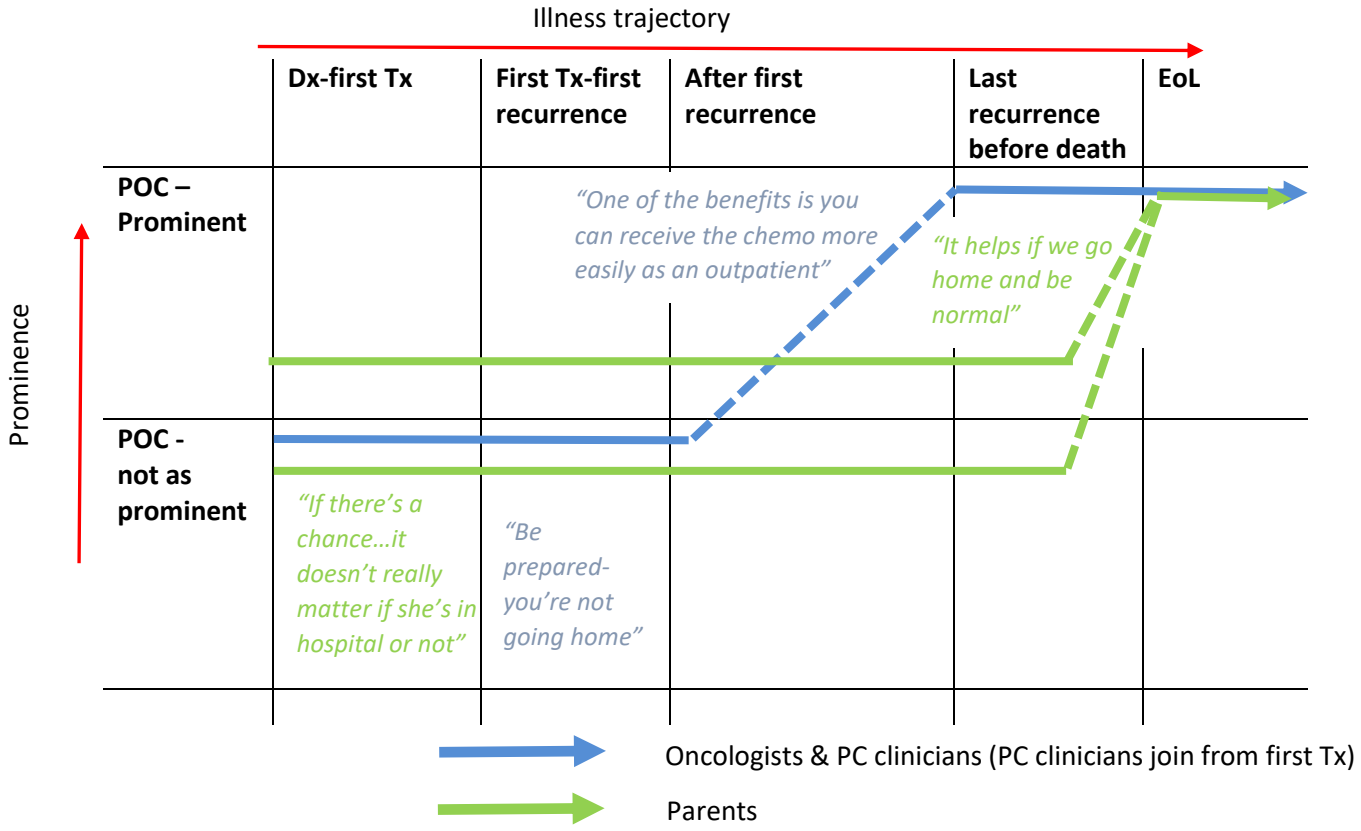


FIGURE 8. THE CHANGING PROMINENCE OF PLACE OF CARE (POC) IN THE PHYSICAL DOMAIN FOR ONCOLOGISTS & PALLIATIVE CARE (PC) CLINICIANS AND PARENTS OF CHILDREN WITH A HIGH-RISK BRAIN TUMOUR THROUGHOUT THE ILLNESS TRAJECTORY



B. SOCIAL DOMAIN

Introduction to domain

The Social domain is made up of four dimensions: School/peer relations, Family relations, Place of care (POC), and Holidays. School/peer relations and Family relations relate to how those social aspects of life enter into the talk around the care and Tx of the child. POC as related to the Social domain focuses more on the social aspects of the locations and what the families gain socially, or not, from the POC, rather than the place as a physical location. Finally, Holidays refer to an extended period of recreation that the child and their families spent out of the home that usually involved socialising together or with extended family or friends. Together, these four dimensions emerging in the consultations represent the social aspects of QoL.

In the following section, School and peer relations and Family relations dimensions are discussed to highlight the changing perspectives of the parents and clinicians with regard to the Social domain. The POC dimension exhibited similar patterns presented in the Physical domain above and so for the sake of avoiding unneeded repetition was not discussed in this section but is summarised in Figure 10 and Table 3. There was insufficient data for the Holidays dimension to undergo comparison across participant groups, hence it is not discussed. See Table 2. in Chapter 4 above for full descriptions of each of the dimensions and Figure 9. in Chapter 4 below for an overview of changing perspectives of School and peer relations and Family relations within the Social domain.

At Dx-first Tx

School and peer relations were a frequent topic of discussion for both participant groups at Dx. For school-aged children and adolescents, school attendance and social relationships with their peers represented what children do. The first quote in Figure 9. suggests this:

> *Dr Louisa Hagan: try to carry on as normal and send her to school*.

Most parents seemed surprised but relieved that their child was expected to attend school in some capacity even through Tx:

> *Shirley Turner: And then she can go to school. At the time -*
>
> *Sally Wiseman: She (stresses next word) <u>can</u> go to school?*
>
> *Shirley Turner: There'll be times when she'll be all right to go to school, yeah…*
>
> *Sally Wiseman: …Oh, she'd love that.*

School and peer relations before progression were encouraged by HCPs and pursued by most parents of school-age children. The social aspect of School and peer relations was particularly emphasised by the oncologists early on:

> *Shirley Turner: If we're doing treatment, our aim would be to get him to school-if we can get him to school a couple of hours in the morning*
>
> *Lin Chen: Uh hmm.*
>
> *Shirley Turner: Because this is a really important age. He's four and a half, meeting his friends, and we'll help you with that…*

First Tx to first recurrence

After Tx commenced, there were more obstacles limiting school attendance for some children, but many parents and HCPs still found ways for children to attend and see friends:

Harold Wiseman: She was pretty much full-time at school throughout (Tx)…she loves it.

Some children attended a hospital school or attended their usual school when Tx and their condition allowed. Friends could also visit outside of school too, allowing an aspect of socialising that some families felt children missed during Tx. However, some children under more intensive protocols who couldn't attend their old schools as much did not enjoy attending the hospital school and missed their old friends. Family relations didn't enter conversations as frequently as School and peer relations at this point before the progression of the disease; friends were still what children and adolescents desired and what seemed a healthy part of life to parents:

Dr Sophie Roberts: How is Arben?

Bora Gashi: he's doing good… I'm happy that he's starting to, you know, plan the weekend with his friends rather than being home and not doing a lot, yes.

PC clinicians who met some families from the point of first Tx also encouraged School and peer relations. The PC team helped families to find ways for their child to still have social relations with peers and attend school when undergoing and balancing the difficulties that came with Tx:

Beatrice Wilson: …maybe the school are doing it in a supportive way…what we try and say is, we want to keep things as normal as possible. So, they're probably saying bring her in because it's normal-

Asalah Kashem: Yeah.

Beatrice Wilson- without understanding she's getting a bit tired. So, we'll give them a call.

After first recurrence

References to School and peer relations for both participant groups diminished after progression of the disease, as did most of the encouragement from HCPs for the child to

attend school and maintain peer relations. For parents, school and their child's peers highlighted their child's difference, as one mother expressed:

> *Asalah Kashem: ...One way she wants to (attend school). Then she doesn't because nobody understands her, she can't do anything she wants…she doesn't want her friends to see her like that as well, even though she misses her friends.*

As the child's condition deteriorated, Family relations came to the forefront for all HCPs and parents. Parents focused on socialising within their extended and immediate family. HCPs encouraged parents to have family time in a place they wanted to be, take holidays, and make memories with their child as one oncologist explained:

> *Hani Singh: Her mum is asking if they stay in UAE for six weeks, will it have any effect on Alia or it's okay?*
>
> *Dr Louisa Hagan: No, no, I think we are in the situation here where the possible gain from experimental Tx is so little in comparison to the gain that Alia will have from being at home with family where she wants to be*

Last recurrence before death

During the last recurrence before death, Family relations dominated discussion for all HCPs. School and peer relations ceased to figure into HCP's talk about what was important. Needless to say, the children's condition often didn't allow for this type of socialising. HCPs also tried to limit clinician presence in the home, so families had time together without intrusion:

> *Beatrice Wilson: Theresa, although she loves us all dearly…she actually wants her home back and she wants her and Anthony to have time with Lisa without us invading it all the time…and I think that includes the on-call overnight, is that right, Theresa?*
>
> *Theresa Kiln: Yeah, definitely don't want to speak to the overnight people.*

At last recurrence before death for parents, Family relations were confined to the immediate family for most children, as one mother described the issues with seeing extended family:

> *Claire Brooke: She needs a bit of a, a decent quality of life –…*
>
> *Bethany Brooke: While we're stuck up here, if she's feeling okay, we're getting her out…If she wants to go and see her family in South London…*
>
> *Claire Brooke: She - she's, you know – it's just so unfair for her.*

Some parents even wanted to limit socialising to the immediate family members at this stage in the illness, and one mother saw family hospice stays as helping limit extended family visits.

EoL

Family relations continued to dominate discussion for both HCPs and parents at the EoL. Family relations remained mostly within the immediate family with families trying to make the most of the time they had left with their child. PC clinicians tried to encourage hospice use to enable quality time as a family, as one PC Clinical Nurse Specialist discussed with the family two days before their daughter's death:

> *Yasmin Blithe: You can stay there (hospice) together as a family, the four of you can stay there…and actually have a proper break.*

FIGURE 9. THE CHANGING PROMINENCE OF SCHOOL AND PEER RELATIONS ('SCHOOL') AND FAMILY RELATIONS ('FAMILY') FOR ONCOLOGISTS & PALLIATIVE CARE (PC) CLINICIANS AND PARENTS OF *SCHOOL-AGE* CHILDREN WITH A HIGH-RISK BRAIN TUMOUR THROUGHOUT THE ILLNESS TRAJECTORY

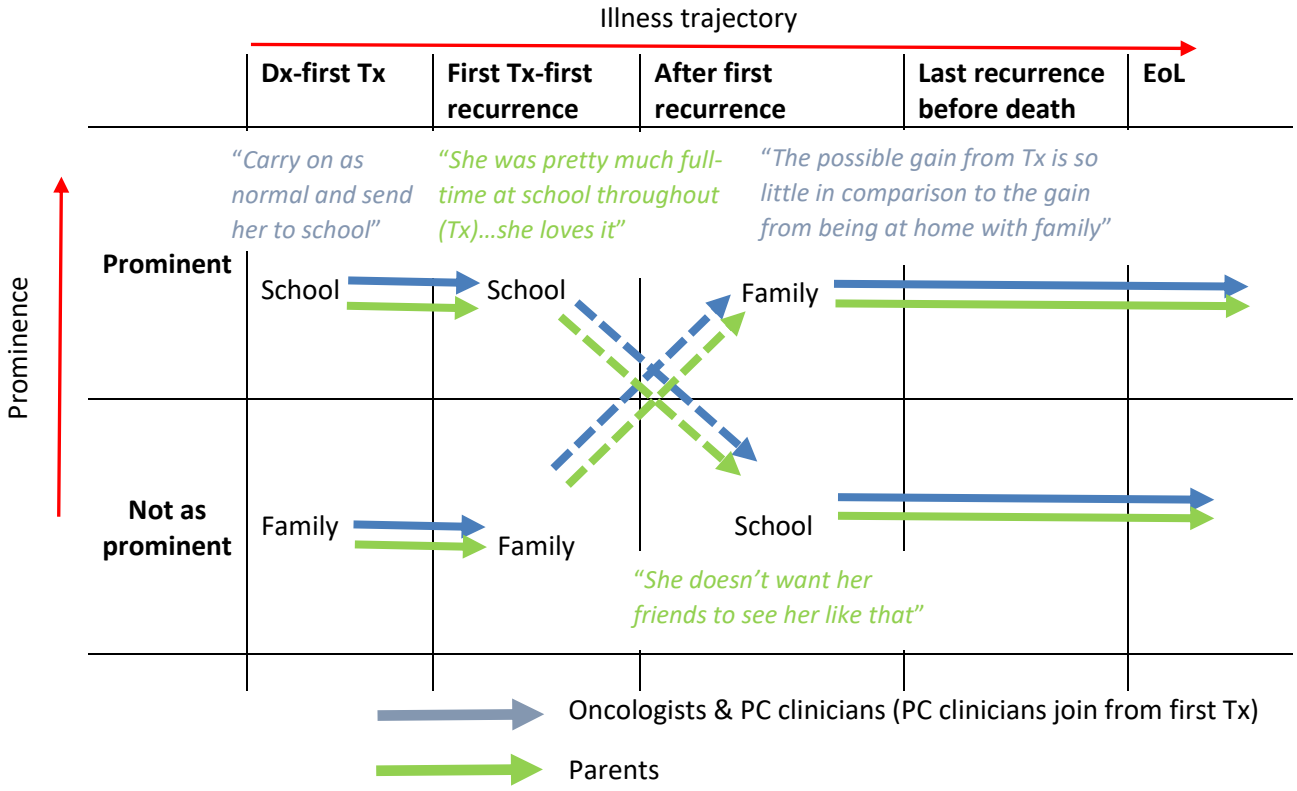
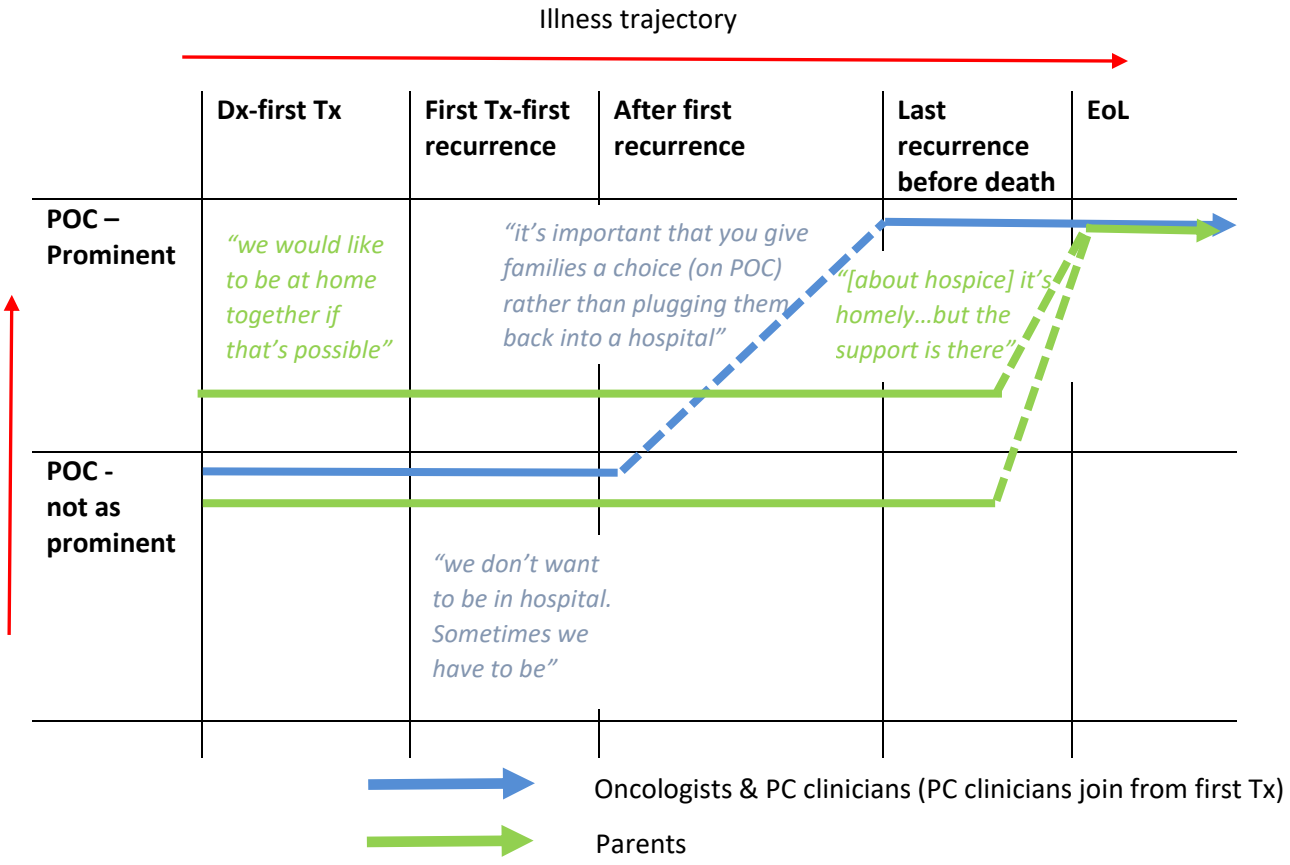


FIGURE 10. THE CHANGING PROMINENCE OF PLACE OF CARE (POC) IN THE SOCIAL DOMAIN FOR ONCOLOGISTS & PALLIATIVE CARE (PC) CLINICIANS AND PARENTS OF CHILDREN WITH A HIGH-RISK BRAIN TUMOUR THROUGHOUT THE ILLNESS TRAJECTORY



C. NORMALITY DOMAIN

Introduction to domain

The Normality domain consists of four dimensions: Construction of normality, Control, Place of care (POC), and School and peer relations. The Construction of normality dimension is taken from Bluebond-Langner's 1996 work with children with chronic illness. Three different constructs of normality are referenced in this domain; i) a 'construct of normality pre-Dx', which is the family's life before illness or that of a 'normal' family without a seriously ill child, a life viewed as 'normal' by society's standards. ii) A 'construct of normality post-Dx', which can also be understood as a 'new normal', where normality has been re-defined and extended to include new aspects of life that result from having a child with a serious illness, such as viewing symptoms or taking medication as normal or routine. iii) Non-normality or 'No longer normal' is the third construct of normality in this domain. As the illness progresses and the child's condition deteriorates, normality can be redefined or constructed to incorporate further changes. But only to a point. There may come a time for a family when redefinition cannot be extended to include the child's condition and all that must be done for her to maintain her comfort.

The notion of the Construction of normality dimension works alongside 'Control', the next dimension in the Normality domain, as a process to reach and maintain that particular construct of normality, the outcome. POC enters the Normality domain in terms of how locations help or hinder families to maintain some Construction of normality in their lives. Finally, School and peer relations, similarly to POC, relate to how school and friends enable the pursuit of normal life.

In this domain, unlike the previous two domains, dimensions were encountered that are changing in nature. The dimensions evolve and new versions are presented in the discussions in which decisions are made about the child's care and Tx throughout the course of the child's illness. The key dimension of the Normality domain is one such dimension. The Construction of normality is presented in this section of the chapter for parents and clinicians. School and peer relations and POC dimensions exhibited similar patterns presented in the Social domain and Physical domain respectively, and so for the sake of avoiding unneeded repetition were not discussed in this section but are

summarised in Figures 12 and 13 below and Table 3. There were insufficient data for the Control dimension to undergo comparison across participant groups hence it was not discussed. See Table 2. in Chapter 4 for full descriptions of each of the dimensions, Figure 11. below for an overview of the key changing perspectives in the Construction of normality dimension.

At Dx-first Tx

The Construction of normality at diagnosis is the same as before diagnosis. The child presented to the oncology team at Dx as a member of a family, school/nursery, and peer group (where applicable). The child is talked about in the same terms as a normal healthy child. Oncologists worked with this Construction of normality and encouraged families to continue attending school, seeing friends and 'carrying on as normal'. Oncologists used a pre-Dx Construction of normality:

> *Dr Louisa Hagan: I mean that is a very big thing to say in the circumstances, but I think she will respond the best if you keep things as normal as possible…if you try to carry on with things as they normally are and she still has her bedtime and she still has to say please and thank you and not be rude-*
>
> *Sally Wiseman: Yeah*
>
> *Dr Louisa Hagan: - and everything else…that somehow tells them that things are okay…*

The Construction of normality pre-Dx, that of a 'normal' child, appeared in the parent's thinking from early on in the illness as a value and as a lens through which life was viewed. This can be seen when a shocked father responds to the news of the Dx:

> *Hodan Omar: He looks just like a normal child.*

First Tx-first recurrence

When Tx began the oncologists started employing a 'new normal', a Construction of normality post-Dx. Symptoms and side effects were included within this post Dx

Construction of normality by the oncologists, and this is what made the construction a new normal for the child, family and oncologists. The parents soon started using this new normal as well. Parents started referring to symptoms as 'normal'. For example, one mother in a discussion with a radiotherapy consultant referred to a potential issue with one of her son's hormones as normal, demonstrating a shift in her Construction of normality post Dx which now included the changed condition as a result of radiotherapy:

> *Dr Shuna Temple: There's also a gland here called the thyroid gland, which can be affected…but you probably know people who have had to take thyroxine…and that's another hormone we'll need to watch.*
>
> *Jolande Marek: Yeah, that's normal.*

PC clinicians who were introduced to some of the families after initiation of Tx also used a Construction of normality post-Dx, reassuring parents for example, that their child's new behaviour since starting steroids or post-surgery, was normal.

After first recurrence

At first recurrence, the oncologist's views diverged from the parents' and PC clinician's views. The oncologist's references to "normality" (the new normality) decreased. The few times oncologists referred to normality they said things like the quote in blue (see Figure 11.):

> *Dr Phillipa Seatter: he's still not back to his normal.*

We take this to mean that not even the new normal could accommodate the changes that had taken place post recurrence for the oncologists. However, PC clinicians still tried to encourage normality, a new normality, throughout the illness trajectory and parents still used their Construction of normality post-Dx and kept on the path of treating their ill child (as they became accustomed to doing once Tx began):

> *Dr Louisa Hagan: Ok so apart from these falls, anything else, any other problems?*
>
> *Hani Singh: Normal headache. It's just a normal, normal rate of the headach*e…

The more the oncologists highlighted the child as no longer normal…:

Dr Bruce Simmons: Now as you know, we've run out of all normal options because unfortunately, high-grade gliomas, there aren't many normal options.

…the further the parents appeared to highlight their child's normality, using their new, post-Dx construction. When asked about symptoms and progression, parents focused on the positives in their child. Even in the face of deterioration parents continued to normalise the child's condition, for example, the quote in green (see Figure 11.):

Rana Jaroudi: (he) sleep normal all night, he wake up, he - normal everything.

Last recurrence before death

At last recurrence before death, for the parents and PC clinicians, the use of a Construction of normality post-Dx increased. Parents and PC clinicians discussed the normality of the child more than any other time throughout the illness trajectory. For the oncologists the use of a Construction of normality (pre/post-Dx) had already dramatically decreased by the last recurrence before death, and it continued to be absent in their discussions. As oncologists were trying to point out the lack of normality, parents emphasised any normality they could, sometimes calling attention to behaviour that one would see in a healthy child:

Rana Jaroudi: …He everything normal…he eating, everything…I don't know what can I do to him, just I pray to him.

At this stage parents continued to accommodate new clinical signs of deterioration within a conception of normal:

Nadia Jaroudi: His breathing –…

Rana Jaroudi: Is normal, it's not just better.

EoL

Only very late in the illness trajectory, just before the death of their child, the parents' view changed and re-aligned with the oncologists' and they too developed a Construction of normality different from the pre or post-Dx Construction of normality. Only now the child

was no longer normal. It was no longer possible for either participant group to view the situation as normal. This coincided for the parents with the child losing much of their social self, such as their communication and/or consciousness, as an oncologist informed a father when discussing radiotherapy for an infant near the EoL:

> *Dr Bruce Simmons: she wouldn't be, you know, Lisa that you know or any, any chance of leading an independent life.*

However, the PC clinicians continued with their Construction of normality post-Dx. PC clinicians continued to focus on how to maintain normality, again, a redefined one, even at the EoL for the families as opposed to the child her or himself. This is illustrated by a PC clinician who tried to maintain some normality of family life even though the child was in a coma:

> *Edith Hutchings: Yeah. Would she – would it be useful to sort of spend time with Sandy (play specialist) coming up with a daily schedule?... Sometimes actually having a schedule helps get somebody back onto the normal …*
>
> *Claire Brooke: It might be helpful*
>
> *Edith Hutchings: daytime, night time thing.*

The change to the construction of non-normality often coincided with an absence of the child's social self; communication and consciousness. As one father put it, normal seems so long ago:

> *Yasmin Blithe: …what was her normal-before all this kicked off and… she got sick, did she go (to the toilet) every day?*
>
> *Theresa Kiln: Yeah.*
>
> *Anthony Kiln: Yeah.*
>
> *Yasmin Blithe: So, you should aim for what's her normal…*
>
> *Anthony Kiln: Normal, normal seems so long ago.*

FIGURE 11. THE CHANGING CONSTRUCTIONS OF NORMALITY FOR ONCOLOGISTS, PALLIATIVE CARE (PC) CLINICIANS AND PARENTS OF CHILDREN WITH A HIGH-RISK BRAIN TUMOUR THROUGHOUT THE ILLNESS TRAJECTORY

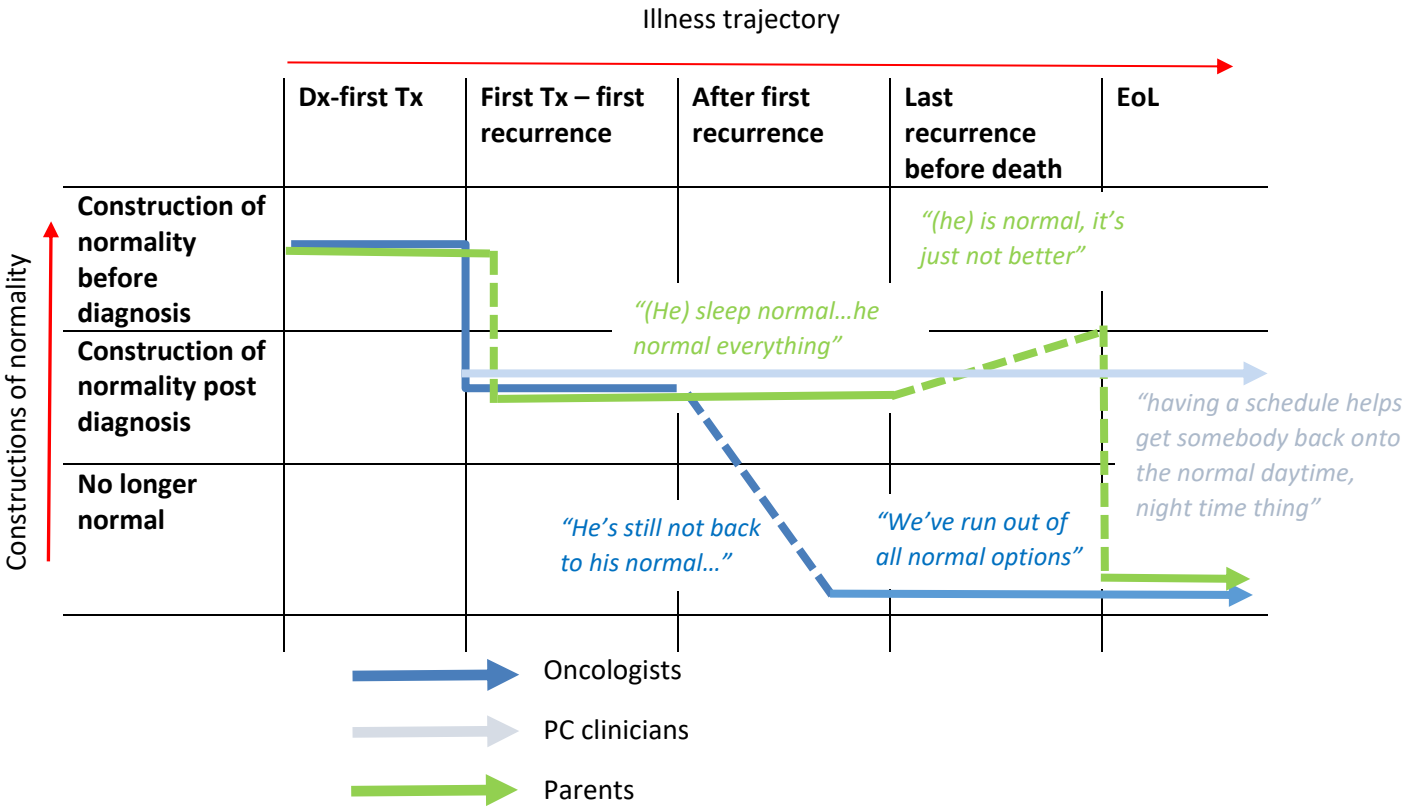


FIGURE 12. THE CHANGING ROLE OF PLACE OF CARE (POC) IN THE NORMALITY DOMAIN FOR ONCOLOGISTS & PALLIATIVE CARE (PC) CLINICIANS AND PARENTS OF CHILDREN WITH A HIGH-RISK BRAIN TUMOUR THROUGHOUT THE ILLNESS TRAJECTORY

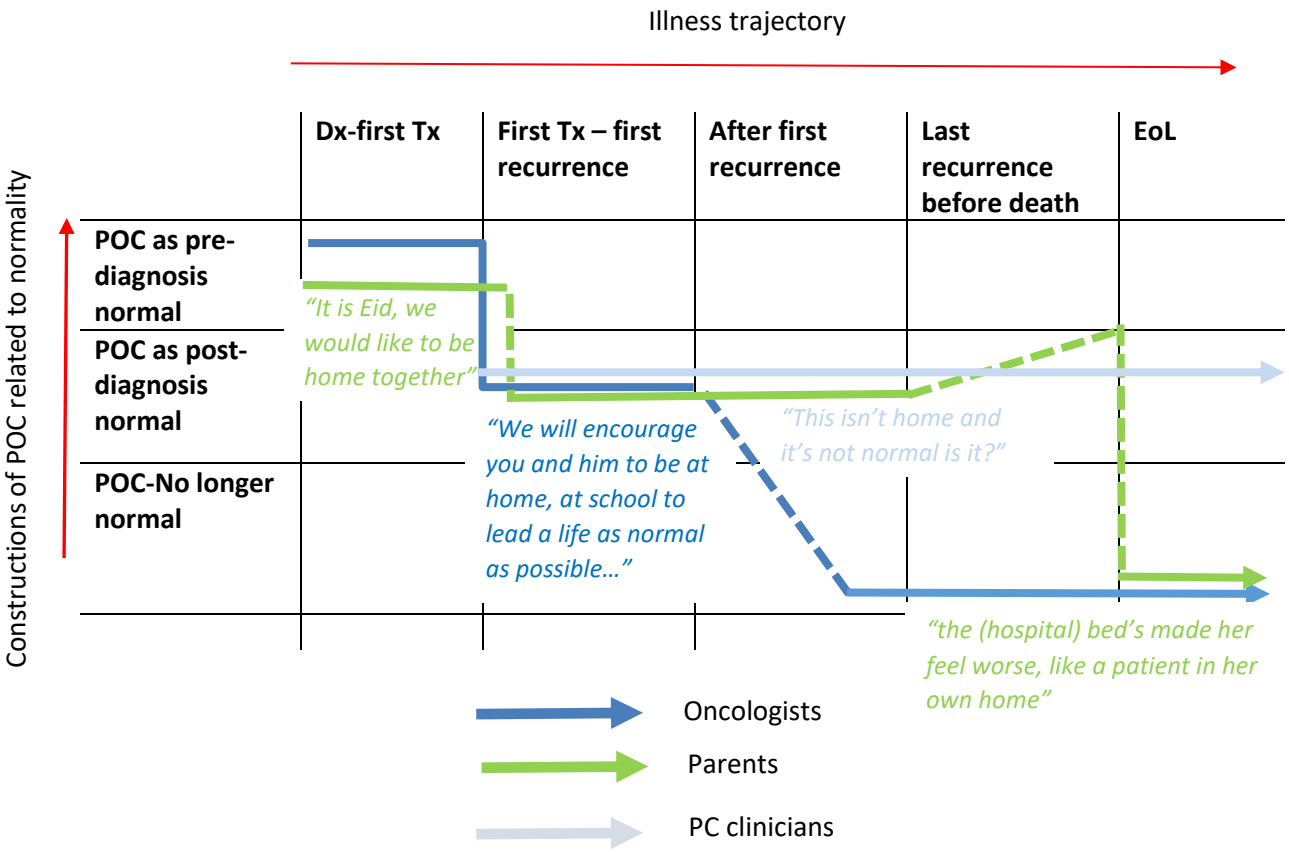
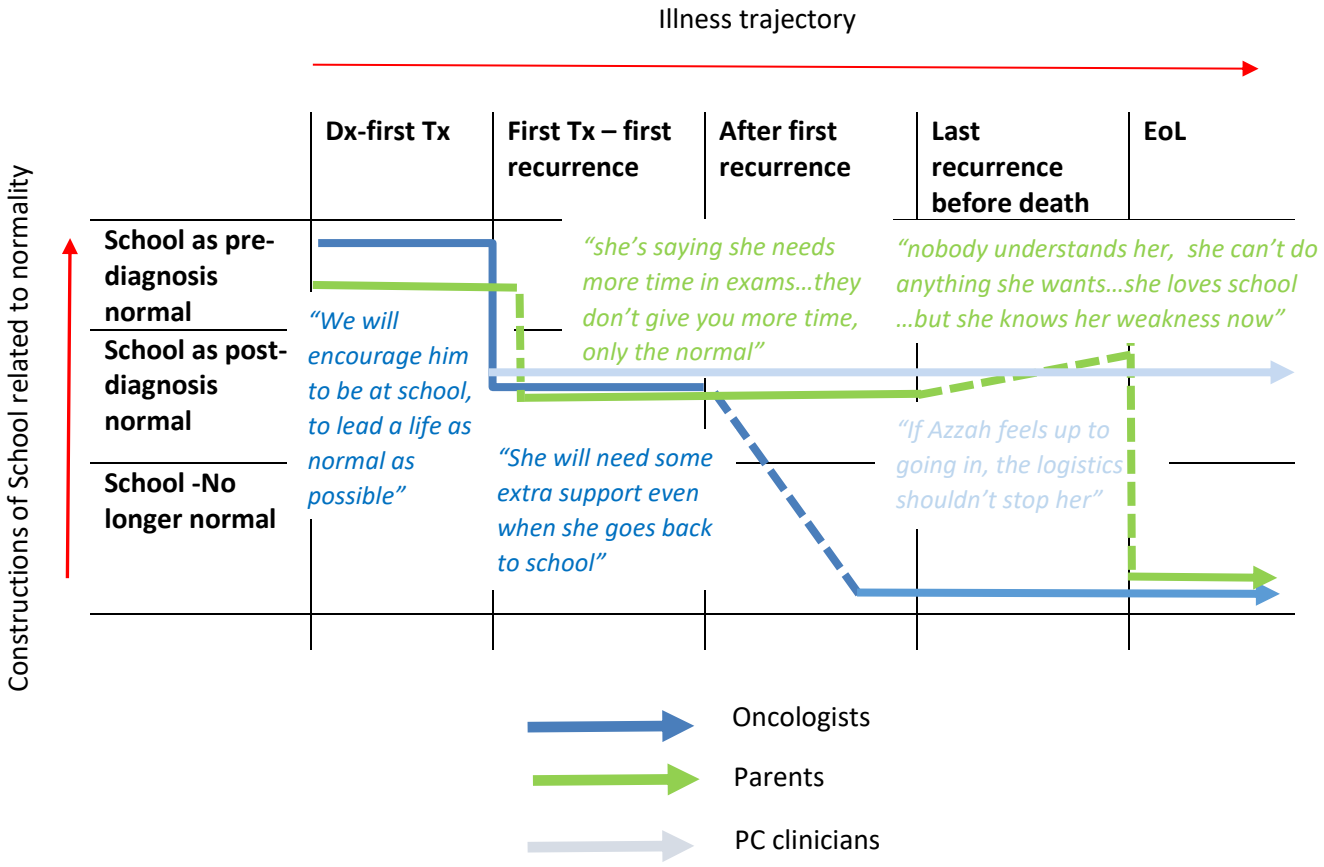


FIGURE 13. THE CHANGING ROLE OF SCHOOL AND PEER RELATIONS IN THE NORMALITY DOMAIN FOR ONCOLOGISTS & PALLIATIVE CARE (PC) CLINICIANS AND PARENTS OF CHILDREN WITH A HIGH-RISK BRAIN TUMOUR THROUGHOUT THE ILLNESS TRAJECTORY



D. FUTURE DOMAIN

Three dimensions comprise the domain of 'Future': 'Construction of the future', 'Wishes', and 'Time'. 'Construction of the future', similarly to the Construction of the normality dimension in the Normality domain, captures the way the participant groups can change their views or reconstruct the future as their circumstances change, altering the horizon of the future and its content. This was found to occur in children with a chronic and ultimately terminal illness (Bluebond-Langner, 1996). The changing construction of the child's future is followed across the illness trajectory along with 'Wishes', which are discussed increasingly throughout the illness and in response to the ever-shortening future. And lastly, 'Time' is seen as a limited and precious commodity. As with the Normality domain, the Construction of the future dimension is dynamic. Rather than just becoming more or less prominent over the course of the illness, it also changes in its content or substance. This section of the chapter focuses on how the Construction of the future dimension evolves in discussions regarding the child's care and Tx, throughout the illness trajectory for the parents and the clinicians. There were insufficient data for the Time and Wishes dimensions to undergo comparison across participant groups and therefore they were not discussed in the thesis. See Table 2. in Chapter 4 for full descriptions of all three dimensions, and Figure 14. below for an overview of the changing perspectives in the Construction of the future dimension.

At Dx-first Tx

At Dx, oncologists talked of an uncertain future that included possible long term (L/T) effects including fertility issues or secondary cancers and tumour recurrence. They also expressed hope for L/T survival and independent living for the children, with better prognoses as articulated by one radiotherapy consultant (see Figure 14.):

> *Sally Wiseman: So, will she be able to get a job…?*
>
> *Dr Shuna Temple: I, I, I…she's going to function…my hope is that she would be able to lead an independent life in the future…but she may require extra support.*

Similar to oncologists, parents' hopes were for survival and even cure, and, for some parents, school attainment. Before recurrence, oncologists still viewed the child, especially those with better prognoses, as one who *might* have a longer-term future, and the current Tx was worth enduring the side effects for this possibility. Even for those children with worse prognoses, Tx was the key to prolonging life and thus was still presented with similar urgency. As one Radiotherapy consultant stressed:

> *Dr Shuna Temple: I don't want you to become worried about this (side effects) because the most important thing is that we get on with her treatment. And if these things (side/late effects) should happen in the future, we will be following her up very carefully and we will deal with them.*

Oncologists encouraged some parents to readjust their expectations about the future successes of their children (e.g. vocational college not a university, and support children may need in school in the future) and parents, as well as oncologists, started to reconstruct the child's future.

Many parents asked numerous questions about the child's future, however the clinician only offered responses reflecting uncertainty. One family discussed the best time to go away on holiday but the oncologist admitted he couldn't answer this:

> *Dr Toby Mansell: But if he has to go, he has to go now… not October (in the future).*
>
> *Hamees Jaroudi: Oh, what's going on in October?*
>
> *Dr Toby Mansell: You're asking me to predict what the future is.*
>
> *Rana Jaroudi: Yeah.*
>
> *Hamees Jaroudi: Oh no, yeah.*
>
> *Dr Toby Mansell: I can't predict it.*

First Tx – first recurrence

After Tx began the oncologists continued to talk about the future in uncertain terms. Oncologists, however, reminded families of children with poorer prognoses that the effects of radiotherapy do often wear off:

Dr Bruce Simmons: …. radiotherapy is a good treatment for this (brain tumour) as in the majority of patients will actually show this clinical benefit and many will actually show the same radiological benefit… But what… you need to be aware of is that unfortunately, in the vast majority of cases, it is a benefit that is not long-lasting. How long it will last varies from child to child but it is something that unfortunately even though when you see such a benefit we know that it's very likely that in the future she'll start to develop the same types of symptoms she had at the beginning. And the benefits of the radiotherapy will wear off.

Children's futures also depended on how they recovered from Tx. Some oncologists expressed hope, even before recurrence, that Tx would provide families with 'a good period of Time' and that they could 'enjoy some Time at home', already implying a limited and reimagined future for the child. Parents adjusted their hopes for the future from the initial Dx consultations with the oncologists.

For many families, a L/T future was more unlikely, and the focus seemed to be more on the present, compared with those who had better prognoses and were more likely to have a longer future.

Sometimes, however, indirectly, oncologists implied some elements of a distant future for children in the study by talking about fertility, university, careers, secondary cancers in adulthood, and follow up clinics that saw the children 'until they were 16', giving the family mixed messages:

Dr Louisa Hagan: …and also with one of these drugs she is at an increased risk of secondary cancers…

Gurey Dahir: Another one?

Dr Louisa Hagan: Usually it's leukaemia, if it is a secondary cancer, it usually turns out to be leukaemia. Usually happens within <u>seven or eight years</u> after you've finished treatment for the first one.

When Tx was well underway, for those children receiving radiotherapy, the oncologists talked more concretely about the extra support children needed in the future post-radiotherapy, at school and in life, thus helping the parent to reimagine the child's future

more concretely. The PC team were introduced to some families after Tx had begun and informed families that the PC nurses could go into schools in the future to educate the teachers on how to best support the child. Again, by doing this the PC team helped to make the reconstructed future less abstract.

After first recurrence

After recurrence, the oncologist's aims and thus the future of the child and family which they envisioned changed into the slowing of further tumour growth and the extension of life with good quality. Tx options tended toward ones with few side effects that had low impact. The sacrifices and endurance of unpleasant side effects for the hope of a distant future made less sense to clinicians after recurrence. As for the oncologists, the future had substantially shortened. The oncologists' earlier more wide-ranging view of the children's future became more narrow. Oncologist's Construction of the future became 'Future as short and with a focus on other QoL dimensions' (see Figure 14.). After the disease had recurred, oncologists and PC clinicians focused on other dimensions in the present rather than in the future. Parents, in contrast, continued to hope for a L/T survival and pursued further Tx but their Construction of the future also now included possible further tumour recurrence and the death of their child. One father repeatedly expressed his worry about further tumour recurrence (see Figure 14.):

> *Hani Singh: (I) worry that in the future if this (tumour) started to appear again*

The same father, later in the consultation, also celebrated his daughter for choosing suitable subjects that would allow her access to university in the future. The following quote emerged immediately after the father had just received news of another recurrence of his daughter's disease:

> *Hani Singh: from the positive side, the school has been supportive to her…and they checked that these subjects are sufficient for future in terms of college and university.*

This demonstrated parents' ability to hold parallel or multiple viewpoints concurrently and their ability to focus on different dimensions of QoL at the same time.

Despite their new short Construction of the future, oncologists sometimes still discussed events taking place in 10/15 years. The child's future fertility and potential secondary tumours were still discussed and continued to give the families mixed messages about a future. However, post recurrence, the uncertainty expressed by HCPs shifted to not *if* they would survive but *how long* that survival would be.

The importance of the family having quality time left was still used by the oncologists and PC clinicians to dissuade families from further aggressive Tx:

> *Beatrice Wilson: When you've got a family, who want to spend as much time- as they would like with their child, it's important that you give families a choice rather than plugging them back into a hospital system.*

Last recurrence before death

At the last recurrence before death period, the oncologists' construction of the future changed again, and the horizon shortened. The future, although still referred to as uncertain, was also referred to (with some confidence) as being very limited now, as one clinician demonstrated in his response to a parent's question about time left:

> *Rebecca Jones: how much time does he have left?*
>
> *Dr Bruce Simmons: realistically, probably something in the order of 12 weeks….*

Any disease-directed Tx focused on drugs with minimal side effects. With this change came advice that the future focused on pain relief and keeping the child comfortable:

> *Dr Bruce Simmons: So, at the moment we don't… think he has any pain, but if he does have any pain in the future, keeping him comfortable…sometimes some of the medicines cause more sedation and we don't want to cause him more problems with medicines.*

Most HCPs encouraged families to enjoy the precious time left with their child at home, or in a hospice, but especially not in the hospital. Oncologists emphasised that the focus of the Tx in the last recurrence before death should be good QoL rather than the aim of slowing or eliminating the tumour:

Dr Bruce Simmons: The success of whether it (the Tx) would be judged on whether or not that responded but also the success must be judged on what quality of life he has. So that we're not giving extra side effects that we think are due to the medicine and not due to the tumour growing.

Many parents continued to hold multiple views about the future of their child until very late in the illness trajectory. For example, in the same conversation, one family talked about what to do with their child's body post-death alongside continuing to enquire into further disease-directed Tx.

EoL

Most parents only moved into the last Construction of the future, 'Future as short/limited', at the very EoL. It was only at this very late point in the illness trajectory that parents' Construction of the future aligned with the clinicians, and parents no longer were able to hold multiple views about the future.

Two weeks before their daughter's death, one PC nurse asked a mother if she had thought about the future. The PC Clinical Nurse Specialist helped them to start thinking about plans and preparing for their child's death and the family seemed ready to discuss their daughter's lack of future:

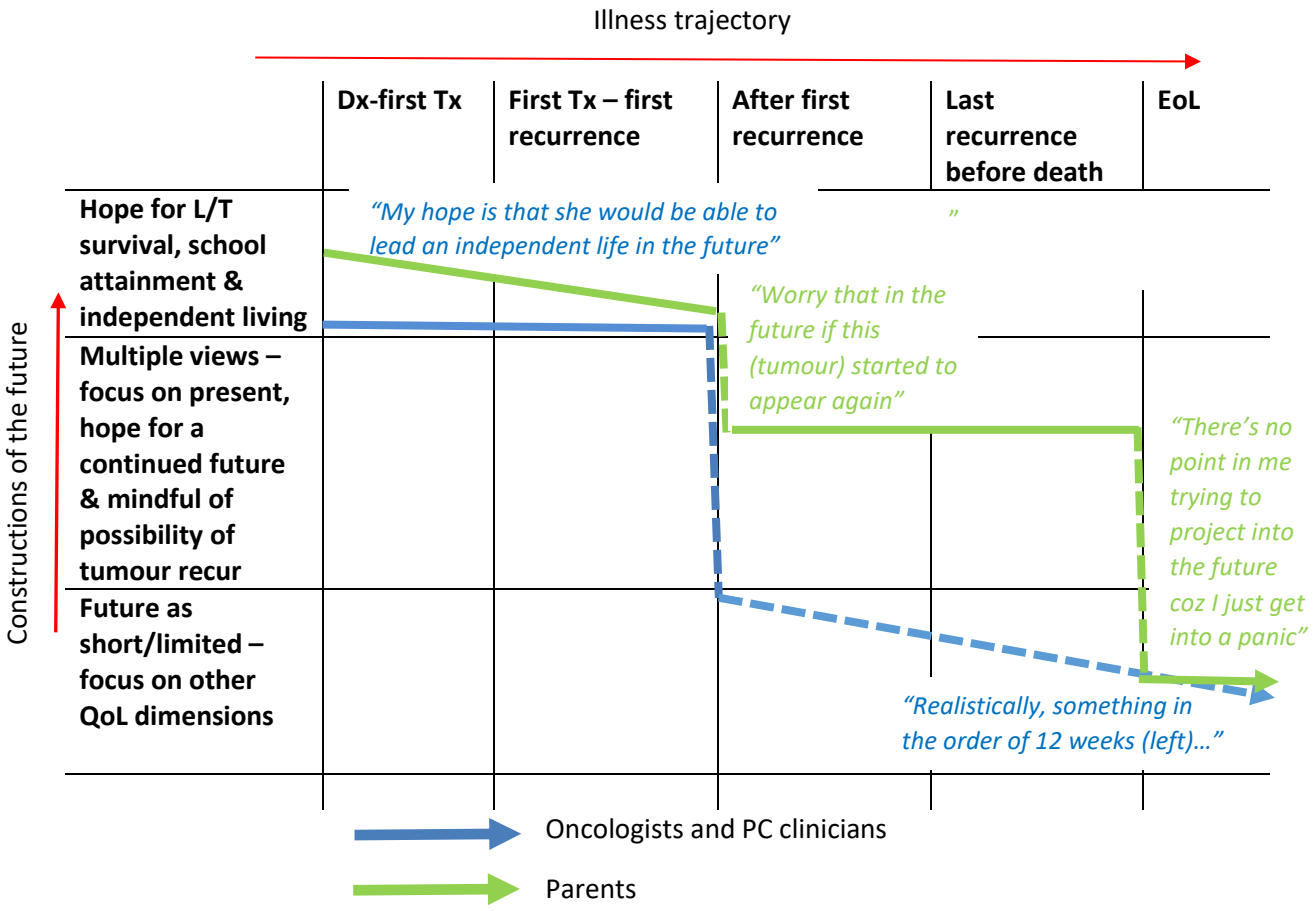
Beatrice Wilson: Are you thinking about the future?

Theresa Kiln: Yeah...you focus on stuff don't you? …You just have to focus.

At this late stage, parents were more able to focus on other QoL dimensions in the present than continuing to look to Tx and hope for L/T survival in the future. The parent in the following quote claimed that focusing on the present helped them to cope with the situation and they still couldn't focus on anything in the future (see Figure 14.):

Claire Brooke: There's no point in me trying to project into the future coz I just get into a panic.

FIGURE 14. THE CHANGING CONSTRUCTIONS OF THE FUTURE FOR ONCOLOGISTS, PALLIATIVE CARE (PC) CLINICIANS AND PARENTS OF CHILDREN WITH A HIGH-RISK BRAIN TUMOUR THROUGHOUT THE ILLNESS TRAJECTORY



4.3 THE DISCUSSION OF FINDINGS

This chapter aimed to explore how QoL entered into clinical consultations in which decisions about the care and Tx of children with HRBTs are discussed and taken. The participants held differing concepts of QoL that changed and adapted to circumstances at different times in the illness.

There were 11 QoL dimensions: Construction of normality, Control, Construction of the future, Disease-related symptoms, Effects of Tx, Family relations, Holidays POC, School/peer relations, Time and Wishes. To illustrate the differences between participants' perspectives of QoL, Chapter 4 presented the key dimension/s within each domain and how these differed between the parents and clinicians over five periods spanning the course of the illness. Because of the limited data on the children's views of QoL from looking at the verbatim transcripts, only the parents' and clinicians' perspectives were presented in Chapter 4. The key differences and similarities (parent-clinician alignment) are discussed below. The clinical implications of the parents' and clinicians' perspectives on QoL are also discussed at the end of 4.3.

Effects of Tx vs other QoL dimensions

At the outset of Tx, the Effects of Tx were not prominent in both clinicians' and parents' QoL concepts and both groups normalised the Effects of Tx. The side effects were seen as something the child and family must endure to reach the potential of L/T survival and therefore other QoL dimensions were 'traded off'.

School and peer relations figured into some family's QoL concepts when the school was possible to fit in around the Tx. School attendance also worked to maintain some normality in the family's lives.

POC, for some parents, served a similar purpose of bringing normality and choice in at a time where normality and choice were lacking. Some families were comforted by choosing to be at home where possible, although other families were reassured by the expertise of the hospital setting.

At progression of the disease, clinicians no longer normalised the Effects of Tx when it was clear to the oncologists that the child would not have a L/T future. Oncologists looked to balance the Effects of Tx with other QoL dimensions and focus moved to Family relations and POC, particularly the home. Parents continued to normalise the Effects of Tx, and were willing to trade off other QoL dimensions to pursue Tx. This finding is related to a '*redefinition of normal'* from Bluebond-Langner's (1996) in which normal is redefined related to clinical events. More and more symptoms and illness-related activities (e.g. physiotherapy) are incorporated into what is seen as normal until interactionally it ceases to work and the intrusion of the illness can be contained no more.

Parents held parallel views from progression; they still hoped for a L/T future for their child and thus pursued further disease-directed Tx, but also held a view that included possible tumour recurrence and therefore were able to focus on making special time for their child, whether at POC or with family.

The balancing of dimensions changed after progression for the parents. Although parents were still able to normalise Effects of Tx, they were able to simultaneously focus on different QoL dimensions depending on which lens they employed or which parallel view they took, depending on the situation. This highlights a fundamental issue in cancer care and PC; that clinicians focus on QoL dimensions that parents aren't willing or ready to focus entirely on at the same point in the illness trajectory.

Multiple views over the course of the disease led to the parents entertaining seemingly conflicting views on aspects of QoL, such as a parent making plans for a funeral but focusing on further Tx too. Recent studies stress the importance of PC being accessed alongside disease-directed Tx, allowing families to occupy multiple and changeable roles for their child (Bluebond-Langner et al, 2017). Parents act as advocates for their children and may need to feel like they have tried every Tx option available to them to help them to live with the decisions they make at this difficult time (Vickers and Carlisle, 2000; Bluebond-Langner et al, 2007; Bluebond-Langner et al, 2017).

Clinician-parent alignment

Although there were clear differences between parents and clinicians, there were some areas of alignment. Before progression of the disease Effects of Tx were not prominent and School and peer relations were prominent for both clinicians and parents. A pre-Dx Construction of normality moved to a post Dx Construction of normality after Tx started, for both parents and clinicians. After progression of the disease, School and peer relations were replaced by Family relations in the Social domain as the most prominent dimension for both parents and clinicians. By the EoL, parents and the clinicians were aligned with regard to most dimensions. Effects of Tx, POC and Family relations were now prominent for both participant groups. The Construction of normality was 'no longer normal' and the Construction of the future realigned to be short and focused on the present for both parents and clinicians by the EoL (see Table 3. in Chapter 4 below)

TABLE 3. OVERVIEW OF MOST PROMINENT & CHANGING DIMENSIONS FOR PARENTS AND CLINICIANS IN EACH OF THE FOUR DOMAINS THROUGHOUT THE ILLNESS

Participant group	Domains	Period in the illness trajectory				
		Dx-first Tx	**First Tx-first recurrence**	**After first recurrence**	**Last recurrence before death**	**EoL**
Parents	**Physical**	Effects of Tx	n/a	n/a	n/a	Effects of Tx
		POC (for some)	POC (for some)	POC (for some)	POC (for some)	POC for all
	Social	School and peer relations	School and peer relations	Family relations	Family relations	Family relations
		POC (for some)	POC (for some)	POC (for some)	POC (for some)	POC for all
	Normality	Pre-Dx Construction of normality	Post-Dx Construction of normality	Post-Dx Construction of normality	Post-Dx Construction of normality	No longer normal
		POC as pre-Dx normal	POC as post-Dx normal	POC as post-Dx normal	POC as post-Dx normal	POC as no longer normal
		School as pre-Dx normal	School as post-Dx normal	School as post-Dx normal	School as post-Dx normal	School as no longer normal
	Future	Hope for L/T future	Hope for L/T future	Multiple views	Multiple views	Future as short
Clinicians	**Physical**	n/a	n/a	Effects of Tx	Effects of Tx	Effects of Tx
		n/a	n/a	POC	POC	POC
	Social	School and peer relations	School and peer relations	Family relations	Family relations	Family relations
		n/a	n/a	POC	POC	POC
	Normality	Pre-Dx Construction of normality	Post-Dx Construction of normality	No longer normal (oncologists only, PC clinicians post-Dx construction till death)	No longer normal (oncologists only)	No longer normal (oncologists only)
		POC as Pre-Dx normal	POC as post-Dx normal	POC no longer normal (oncologists only)	POC no longer normal (oncologists only)	POC no longer normal (oncologists only)
		School as pre-Dx normal	School as post-Dx normal	School no longer normal (oncologists only)	School no longer normal (oncologists only)	School no longer normal (oncologists only)
	Future	Hope for L/T future	Hope for L/T future	Future as short	Future as short	Future as short

Key

Physical = Effects of Tx and POC
Social = School and peer relations and Family relations
Normality = Construction of normality
Future = Construction of the future
n/a = no dimension prominent in the domain for that period in the illness

QoL as a dynamic concept

As illustrated above, although QoL was a concept that had some alignment between parents and clinicians, QoL for both participant groups was dynamic in nature. This supports previous research that individuals' evaluations of their QoL may change over time (Ferrans, 2004; Hinds, 2010). Physical and Social domains contained univocal dimensions that changed in prominence, becoming more or less prominent or important throughout the illness trajectory. Normality and Future domains, however, changed in nature and response to the family's changing circumstances.

This finding aligns with previous research that concludes that QoL is a multidimensional construct (Matza et al, 2004) but is taken further in the current thesis by adding important insights into how those dimensions behave over time as well as into the lack of homogeneity between certain dimensions. Both Normality and Future domains contained changeable elements; e.g. in the Normality domain, the Construction of normality started off as a Construction 'pre-Dx', and after Tx started progressed to the new normal 'Normality post-Dx', then at different points the two groups moved to the final Construction of normality, 'No longer normal'. What the different Constructions of normality included changed also. At first, school was included in the Construction of normality; then, at progression, school was no longer equated with normality but conversely highlighted differences, e.g., a 'hospital' school, missing exams, looking different in appearance to other children or being physically weaker than peers. Past research has interpreted QoL as a striving for normality or a new normal (Clarke-Steffen, 1997; Deatrick et al, 1999; Van Schoors et al, 2018; Beecham et al, 2019). This interpretation aligns with this thesis, but also develops the literature through the finding that clinicians and parents all have different concepts of what constitutes Normality for their child with an HRBT. The concepts also change and evolve differently throughout the illness trajectory. Families can redefine or readjust what Normality means to them. This is similar to what Bluebond-Langner found in children with cystic fibrosis back in 1996 and Bluebond-Langner and colleagues found in 2017 in children with LLC/LTI generally.

The Construction of the future dimension within the Future domain also included different elements. At Dx parents hoped for L/T survival and school attainment. Then at

progression they took on multiple views, still hoping for L/T survival but realising that the parent's future also contained possible further tumour recurrence and death. Having a future is usually definitive of childhood and so this future is difficult for parents to lose (Bluebond-Langner, 1978). Past research has found that individual perspectives regarding the future were not shared between parents and HCPs (Verberne et al, 2021). Verberne et al (2021) found that some parents reported that HCPs had not been open to exploring the future or answering their questions, mainly because of prognostic uncertainty. The current study shows that the different perspectives about the future *were* shared in different and sometimes indirect ways between the two groups, but that similarly to Verberne et al (2021), some clinicians hid behind uncertainty, which limited discussions about the future. It was only at the very EoL that most parents aligned with clinicians and no longer held parallel views, allowing the other QoL dimensions to come to the forefront over Tx of the disease. As the child's function declined progressively and symptoms increased, renormalisation and redefinition of the future adapted (up to a point) to these changed circumstances.

Addressing the issues in the QoL measurement literature

This evidence of the dynamic nature of QoL has implications for QoL measures and tools. As stated elsewhere in this thesis, HCPs often depend on QoL and health-related quality of life (HRQoL) measures to compare outcomes and reach decisions on a child's care and Tx (Eiser and Morse, 2001c; Yock et al, 2014), including participation in clinical trials and using experimental therapies. HRQoL measures are also used to evaluate nurse-led interventions (Madden et al, 2010). Our findings about the dynamic nature of participants' views of QoL may present a challenge to the cross-sectional comparison of QoL measurements.

There has been a recent proliferation of HRQoL instruments, yet questions have been raised about the use of HRQoL measures in cancer in children (Anthony et al, 2017) and more specifically in paediatric BT populations (Hinds, 2010). Paediatric oncology QoL instruments are deficient concerning 'content validity', that is, they fail to attend to what is important to children with BTs (and their families).

Results illustrate that what is included in our QoL concepts is subjective. Preferences of parents and clinicians vary between participant groups and over time. Dimensions of QoL vary in type and nature. To operationalise these QoL concepts into an instrument to measure QoL would require more than a single, static measure. Also, concurring with Eiser and Morse (2001a) and in keeping with the interactionist perspective, this thesis recommends using the views of all three participant groups to understand the QoL of the child. Also, more recently, Avoine-Blondin et al (2018) recommended that QoL be evaluated together with the family, child and clinician as a team. As Landgraf and Abetz (1996) argue, it takes children until age 9 or 10 to report on abstract concepts such as their self-esteem, and thus a holistic approach to understanding a child's QoL, including other groups involved in their care, can aid in the clinicians' understanding.

Chapter 4 presents further problems for the reliance on HRQoL measures as a basis for the improvement of services. Services and interventions for BT patients and survivors need to be developed with an accurate understanding of the family's experiences and situation. Models of care and interventions should be guided by what has been learned in this thesis about the subjective and changing dimensions of QoL. Fakhry (2013, p.438) recommended: '*in the terminally ill population, it is even more important to make evidence-based decisions to promote HRQoL, as this becomes the only true measure of the impact of medical intervention*'. The touchstone for determining the adequacy of interventions and services lies within the family's interpretation of the situation in which they find themselves- not what that situation is 'objectively', but what they make of it as they adapt and make meaning in the circumstances of which illness has become an overwhelming part.

Clinical implications - from the parents' and clinicians' perspectives on QoL

The scheme of the four domains is used to frame the discussion of the implications for clinical practice.

Physical domain

Not surprisingly, the study found that clinicians quite understandably change tack regarding side effects at progression, when the risk/benefit analysis of aggressive Tx changes. Oncologists spent time initially getting parents to expect and endure side effects from Tx. Previous studies have found symptoms to take on different meanings at different periods of the illness (Woodgate et al, 2003; Bluebond-Langner, 1978). At progression, clinicians tended to steer families towards options with fewer side effects. However, some parents are often willing to accept these side effects, some seeing side effects as evidence that the Tx is working (Bluebond-Langner, 1978).

This change in perspectives, however, has the potential for creating misalignment between clinicians and parents. At this point, when clinicians highlight risks of further Tx as a reason to forgo certain options including continuing disease-directed therapy, some parents will push back emphasising the lack of side effects from Tx and/or the stability of the child's condition.

The divergence of viewpoints at progression has implications for maintaining relationships. These consultations at progression are highly complex and emotive, and clinicians often have to respond and act quickly. Clinicians must be supported on how to deal with this complexity.

Findings from this thesis could stand as a first step towards the enhancement and improvement of communication training for clinicians. As noted in a recent editorial in the journal Pediatric Blood and Cancer, the 'reflection' needed for managing complex cases and ambiguity in consultations can be enhanced through working with a trained facilitator and implementing reflexive practice into training curricula (Rainusso & Frugé, 2021).

The theoretical findings from this thesis can be translated into the practical curriculum in two ways: through primary medical communication training, and continuing medical education (advanced communication courses). Communication training courses would deal with reality and situations that the professionals attending have to deal with in their day to day practice. Simulation-based communication training could provide an opportunity to present the principles emerging from this study: managing a triadic interaction with differing priorities and views and the consequences of these differing

views that can arise, the different stages or periods in the illness trajectory, and how views can change at different point, and the content of what changes in the different participant groups' views. Professionals attending these courses could experience these findings through education, which will equip them to be able to recognise the misalignment of views between parents and clinicians that may occur and to practice ways of responding to and reducing misalignment through clear communication and acknowledgement of these differing views in a safe and controlled learning environment (that being an educational environment of a training course). The training course offers a space to practice difficult conversations that may arise in the consultation so professionals can build techniques they can apply in practice in the consultations.

Social domain

Clinicians and parents began with similar priorities regarding the Social dimensions, and these changed in similar ways at similar points in the illness. Children, however, especially older children, were more interested in school than their parents until later in the illness trajectory. The communication training courses available should include a child/parent/clinician scenario and mirror the changing priorities of the different participant groups in the interaction. For example, a parent might focus on Tx but a child might want to balance school/friends, particularly in the case of older children for whom the school may still be a large contributor to some normality in their life. The facilitator could run through real examples of how the clinician can juggle the varying priorities, listen to each participant group, and come up with a solution that meets everyone's needs.

Normality domain

The thesis highlighted the likely times in the trajectory when clinicians and parents' views of normality change (first recurrence and then at EoL) and the consequences of these changes for discussions of Tx options (what the clinician will want to offer and what the parent will ask for). Disalignment was observed between oncologists and parents concerning the child's condition after the first recurrence of the disease and up until the

EoL. These differences in the construction of the child's condition may in part be due to the differences in the Construction of normality. The normalisation of symptoms and side effects by the families can help HCP to understand why parents continue to seek high-dose, intensive Tx up until very late in the illness trajectory against HCP advice. Parents were more willing to continue aggressive Tx post-recurrence because they had normalised the presence of burdensome symptoms in their lives. This is a potential adverse consequence of normalisation from a palliative care perspective. Requena et al (2022) stressed that normalisation of symptoms '*is a pervasive barrier enacted by all involved in caring for children with advanced cancer'.* Authors acknowledged that the normalisation of symptoms resulted in inadequate management of side effects and symptoms and therefore reduced the alleviation of symptoms including pain. The authors concluded: *'Strategies to overcome normalization are critical to ease child distress*' (p.548). Normalisation is a process that is therefore both functional and dysfunctional, depending on one's perspective.

Normalisation is not connected with a change in underlying values, though. Clinicians can maintain relationships with parents when the two groups differ over their views on the burdens of Tx. Understanding the basis of the disagreement is potentially constructive and helpful for clinicians. These scenarios can be implemented into communication-training for clinicians, again, in order to practice ways of dealing with these difficult scenarios when disalignment can occur between families and clinicians. This biomedical bench research, as Gulbrandsen et al (2022) analogise it as, although not solving the impasse that often results in these emotive consultations, is a necessary first step in understanding how to tackle these difficult conversations in an educational context.

Future domain

This study highlighted the ability of parents to hold multiple or parallel viewpoints. Medical training courses could support clinicians in identifying the times when parents seem to be holding contradictory preferences. Facilitators could educate clinicians on the parents' ability to hold multiple viewpoints at once and change focus on QoL dimensions depending on which lens the parent applied. Facilitators can help professionals not only

to learn how to support both viewpoints and recognise manageable alignment and disalignment but also to understand when the conversations become unmanageable and the clinician needs to acquire further support when dealing with a family (through second opinions or the courts) as is needed in some extreme cases of misalignment. Professionals attending the courses could learn how to plan for best and worst scenarios.

4.4 CHAPTER CONCLUSION AND TRANSITION TO CHAPTER 5

This chapter has shown the complex ways in which QoL enters into care and Tx discussions about children with HRBTs. Chapter 4 has presented the perspectives of two of the three participants in the consultation, parents and clinicians, and followed them over the course of the illness. The participants held differing concepts of QoL that changed and adapted to circumstances at different times in the illness.

A gap in the QoL literature and our findings thus far is the QoL perspective of the children themselves, taken from their own voice, in real-time clinical consultations. In the next chapter principles and practices from discourse analysis (Antaki, 2008) are used to examine the child's voice more closely in these interactions. A focused study of one of the adolescents from the study is used to understand how the child's voice emerges in the exchanges between the participant groups when dimensions of QoL are discussed.

CHAPTER 5 – THE CHILD'S VOICE ON QUALITY OF LIFE ISSUES IN CLINICAL CONSULTATIONS

5.0 CHAPTER OVERVIEW

Researchers have emphasised the importance, indeed, the necessity, of understanding the child's perspective on QoL given the subjective nature of QoL (Eiser, 2004; Eiser and Varni, 2013; Huebner et al. 2004; Beecham et al, 2017 [upgrade]). Without understanding the child's perspective, the decisions made in consultations that involve the child may fail to respect what is important to the child. However, little is known about the child's perspective, particularly at the EoL (Hinds, 2010).

It has been found that children's verbal contributions tend to be a small fraction of the total duration of a consultation (Stivers, 2001; Cahill and Papageorgiou, 2007; Miller et al, 2014). Other research, however, argues that a focus on the amount of speaking misconstrues and underestimates the child's contribution to the consultation. Such research suggests that appreciating children's and young people's participation in consultations requires its own unique approach (Stivers, 2001; Clemente, 2015; Kelly et al, 2017; Weaver et al, 2015; Tates et al, 2002a).

"Listening" and "hearing" does not apply literally to the child's voice in consultations. Thus, in order to obtain this crucial child/patient perspective on their own QoL, the complexities of eliciting a child's perspective from the consultations in order to identify a child's concerns about their QoL had to be dealt with. To hear the child's voice in the consultation, it was found that it was not only necessary to use interactive methods to appreciate the children's participation and achieve an understanding of how the child's voice emerges in triadic[3] conversations about QoL, but it was also necessary to refine those approaches to fit the unique character of the triadic process of paediatric clinical consultations. In this chapter, a close analysis of conversations in a single case is used to provide the opportunity to demonstrate the various ways in which a child can participate in a consultation when QoL is discussed and from these, identify the child's views of their

[3] Triad in this chapter refers to three or more participants in the group

QoL. This provides the basis for essential first steps for documenting children's own views on QoL issues and applies them to the limited data which I have from them in this thesis.

The chapter is divided into five sections:

Section 5.1 explains the background to the chapter and an outline of the methods.

Section 5.2 explores the child's participation and involvement across the QoL dimensions in the consultations.

Section 5.3 presents the Discussion of the findings.

Section 5.4 presents Conclusions from the chapter and provides a transition to Chapter 6.

5.1 BACKGROUND/METHODS

Background

To briefly summarise and review the literature described at more length in Chapter 2, while the participation of children and young people in clinical consultations has been recognized as important for a number of reasons both ethical and clinical (Royal College of Paediatrics and Child Health, 2000; American Academy of Pediatrics Committee on Bioethics, 1995), there is a lack of consensus in the literature as to what child participation involves and what form it takes or should take.

A number of medical studies of child participation in consultations have used observations of real-time interactions in consultations, focusing on and measuring child participation by percentage cover of the speech in the consultation (Miller et al, 2014; Olechnowicz et al, 2002). More recent and nuanced studies conducted to explore child participation in consultations have moved from a priori concepts of participation to ones empirically derived from children themselves. These more recent studies have revised previous understandings of participation and, using broader views of child participation, have indicated a variety of participatory roles employed by children (Ruhe et al, 2016a; 2016b; Kelly et al, 2017 and Weaver et al, 2015). However, amongst other limitations,

these more recent studies largely used interview data which depended on the recall of the participants.

Research based upon analysis of recordings of the consultations has also had shortcomings. Often such research focuses on one to one or dyadic interchanges (Tates and Meeuwesen, 2001). However, recent research stresses that paediatric decision-making should be analysed as a triadic process (Bluebond-Langner and Langner, 2021) and calls for more observational research (Quaye et al, 2019), specifically exploring how parents and clinicians can promote child participation in meaningful ways (Clemente, 2009). Stivers (2001) found in a study of well children in US pediatric (= UK GP) consultations that:

> *the child's participation is the product of an interactional negotiation among the physician, the parent, and the child. In focusing entirely on the outcome (i.e., who eventually answers questions and provides information) researchers may miss the process that actually includes children and treats them as consequential parties in the interaction and may further miss the critical differences in participation across activities within the encounter.*

The need for triadic, observational study of child participation is thus well supported.

Methods

A single case, rich in different forms of participation by the child, was selected to explore the child's voice. Alia Singh was a 14-year-old girl from UAE with a high-grade glioma that had progressed twice at the point of study entry. The 77 QoL excerpts from 23 transcripts from the original query used for the analysis in Chapter 4 were used for the exploration of the child's voice for this Chapter. The coding scheme for the child's voice exploration was developed using inductive and deductive terms and was applied to the 77 QoL excerpts (See Table 4. in Chapter 5 below). This resulted in smaller 'participation excerpts' which became the basis for the analysis for Chapter 5. Constant comparison (Glaser, 1965) and discourse and conversation analysis (Antaki, 2008) were conducted on these excerpts and patterns were noted on the codes that emerged and who was

involved in the interactions. Figure 6 in Chapter 3 provides the procedure overview of the development of the excerpt booklet and data analysis.

TABLE 4. CODING SCHEME FOR CHILD'S VOICE EXPLORATION

Coding Category	Codes	Type/Source	Description
Child's participation	Non-verbal	A Priori/Literature - Modified from Ruhe et al (2016b)	The child participates in the consultation through non-verbal responses.
	Verbal		The child participates in the consultation through verbal (English) responses.
Subject of conversation	Decision	A Priori/Literature - Modified from Ruhe et al (2016b)	The child participates in actual decisions being made (in English).
Child's role	Child as translator	Inductive	The child acts as a translator for a parent.
Child's input	Prompted	Inductive	The child speaks in response to another participant's prompts, e.g. an utterance such as 'hm?', repeating the child's name or asking a question in a different way to prompt a response.
	Child-initiated	Inductive	The child initiates a sequence of conversation or joins in the conversation without being asked.
Characteristics of the child's responses	Reformulations	A Priori/Literature- Lehtinen (2005)	Restatement of clinicians' presentation in own words.
	Continuers	A Priori/Literature- Lehtinen (2005)	Words or gestures acknowledging what the clinician has said which allow the clinician to proceed.
	Questions	A Priori/Literature- Lehtinen (2005)	Any question asked by the child.
	Personalisation	Bluebond-Langner et al (2021)	Any question or statement made by the child which applies general or population-based information to the speaker's own, unique situation.
	Responds "*I don't know*"	A Priori/Literature - Modified from Clemente (2009)	When the child responds "*I don't know*" to a question.
	Solicitation of an answer	A Priori/Literature - Modified from Clemente (2009)	When the child seeks corroboration of their given answer from a parent or seeks the whole answer from the parent.
	Speaks in English	Inductive	The child speaks in English.
	Speaks in Arabic	Inductive	The child speaks in Arabic, directed to the mother or father.
Other participants' contributions	Adult states child's views/opinions/desires	Inductive	The parent or clinician states the child's views, opinions or desires.
	Adult reports the child's symptoms/physical condition	Inductive	The parent or clinician reports the child's symptoms or physical condition.
	The parent encourages the child to speak	Inductive	The parent encourages the child to speak, for example, to answer the clinician's question or to give more information in the child's own words.
	Parent answers for the child	Inductive	The parent answers a question directed to the child.
	Clinician answers for the child	Inductive	The clinician answers a question directed to the child.
	Clinician question directed to the child	Inductive	When the clinician explicitly directs his/her question to the child.

Coding Category	Codes	Type/Source	Description
	Clinician question directed to the parents	Inductive	When the clinician explicitly directs his/her question to the parents.
	Clinician question, recipient unspecified	Inductive	When the clinician has no clear intended recipient for his/her question.

5.2 EXPLORATION OF CHILD'S PARTICIPATION AND INVOLVEMENT IN THE CONSULTATIONS

Setting the scene

Alia Singh attended consultations conducted at different times by three different clinicians: most (17) were with her primary oncologist 'Dr Louisa Hagan', three were with a surgeon 'Mr Kieran Williams', and three consultations were with an oncology consultant at another institution, 'Dr Bob Shaw'. Her mother 'Mona' was present during all consultations. Almost all of the mother's speaking took place in Arabic and was directed to Alia's father 'Hani' or to Alia herself. In 8/23 consultations Alia's father was not present and so the task of translating the clinician's speech for the mother and the mother's speaking for the clinician fell to Alia.

When the father was present, he was the translator to and from Alia's mother. Sometimes when Alia spoke in Arabic to one or both of her parents her father then translated for the clinician. This is noteworthy since both the oncologist and Alia's father regarded Alia's English as excellent, the father saying at one point that her command of the language was better than his.

Most of the conversation during the consultation took place between the clinicians and Alia's father. This was true when QoL issues were in play as well. Nevertheless, given the perspective used in the thesis, Alia's participation and contribution are not regarded as proportional merely to the amount of time that her speaking occupied. For example, attention is given to both verbal and non-verbal communication (Ruhe, et al, 2016b), prompted versus unprompted speaking, making space for a parent or clinician to answer, being involved in a decision (Ruhe et al, 2016b), asking and answering questions (Lehtinen, 2005), seeking corroboration, and acting as a translator (see Table 4. In Chapter 5 for the full list of codes and sources).

In addition, some elements of Tates et al's (2002a) interactional sociolinguistic approach were applied in looking specifically at how the adults displayed their roles and identities in the consultation. In the institutional context of a paediatric consultation, I observed this mainly in the form of patient-centred or child-centred behaviours on the part of the adults. Tates et al (2002a) refer to child-centred behaviours and supportive or non-supportive adult behaviour. In the thesis child-centred behaviours were observed, these behaviours

included: emphatically directing speech to the child and meta-communication in the form of urging the child to speak for herself and also admonishing parents not to speak for the child. Thus, I saw in the language of the consultation clinicians and parents enacting their roles in the form of supporting and encouraging the child's participation.

A. EXPLORATION OF ALIA'S PARTICIPATION IN THE INTERACTIONS

Verbatim transcripts of consultations are used here to examine what a child actually did do during consultations. These are the first steps toward showing that and how children can have an impact on consultations far greater than what might be suggested by the amount of time they spend speaking in consultations and toward illustrating ways to assess that impact and "hear" the child's voice.

Specifically, this section looks at child-initiated speaking, the child's questions to the clinician, the child's response to prompts to speak including questions directed to the child, and the child's non-verbal communication.

Alia-initiated speech

Most of the Alia-initiated speech (10 of the 15 total Alia-initiated speech references) took place when Alia's father was absent. The following excerpt illustrates how Alia could offer her opinion, disclose information and ask questions in a conversation with an oncology consultant without being invited to give her opinion or join in:

Dr Bob Shaw: So if the blood sort of flows - when you have a cut it seals it up. Sometimes this sort of sealing of blood can happen in the vessels and then it blocks the vessels and then you sort of get a swollen, painful vein.

Clinician is informing Alia about the possible side effects of a drug

Alia Singh: So you mean it stops them and things like altogether?

Alia-initiated speech -to check she's understood what the clinician has said

Dr Bob Shaw: Yeah, all the blood clumps together in one clot, it doesn't move anymore.

Alia Singh: (speaks but not in English for less than 5 seconds).

Mona Singh: (speaks but not in English for less than 5 seconds).

Alia Singh: So the first one this time is like that here, not much. So like the main side effect is my back, for the pills that I took on-

Alia-initiated speech -to describe her symptoms

Dr Bob Shaw: Back pain?

Alia also interjected into conversations where her father was present too:

Dr Louisa Hagan: And I can't give her temozolomide because if I give her temozolomide I would make her sick.

Hani Singh: (speaks but not in English for 5-10 seconds)

Alia Singh: I took it before.

Alia-initiated speech

Dr Louisa Hagan: I know, but your, your, your count is so fragile…

Alia's Questions

Some of Alia's contributions in the consultations took the form of questions she asked of the clinicians. Nine of the 10 questions Alia asked during these consultations were when her father was absent. Alia's priority to continue with her schooling emerged during interactions with her mother and the clinician. Although Alia expressed that she felt unable to attend school full time, she still wanted to be able to attend as much as her illness allowed her. Alia stressed that her teachers needed to better understand her need for flexibility at school. Alia asked if the oncology consultant would write to inform the teachers about her needs:

Alia Singh: And (do) you want to send anything for school because I'm starting on…

Minor question asked

Dr Louisa Hagan: Okay, yeah could I have the name of the school and stuff.

School was the subject of several of Alia's questions. However, Alia asked more major questions on the Effects of Tx dimension, specifically about the potential side Effects of the Tx. She sought clarification about a spot on a scan a clinician showed Alia asking if the side effects would go over time "*and like does it go through time?*". She also sought reassurance that the latest deterioration was not the tumour (see excerpt below). Alia asked the question continuing with her mother's refrain (as translated by Alia; see below) of checking if the clinician was sure whether the recent deterioration was the tumour or not:

> *Dr Bob Shaw: Okay. That is my expectation. But, like everything in life and you know it better than I do, it's not a guarantee and it still may be that actually the problem is related to the tumour and it may not have responded to treatment. But at the moment my hope is this is only inflammation which will settle with a little bit of infusion very, very quickly.*
>
> *Alia Singh: So we are not still sure if it's a tumour?* } Major question asked without being invited
>
> *Dr Bob Shaw: Well, we would only be absolutely sure if you would undergo another operation. But we think in this area, after the things you've gone through, to put you through another operation just to prove A or B is not fair.*

Prompted speech

There were 27 references across the consultations where Alia's speech was prompted by a clinician or parent. Most of the prompted speech (22/27 references) occurred when the father was present. A prompt could be simply a clinician's utterance such as 'hm?', repeating the child's name or repeating a question or asking a question in a different way to prompt a response.

Some questions were more straightforward and concrete such as: 'How old are you?' or 'Have you been to school?' Alia generally responded and answered more promptly, elaborating on her answers, than the more judgment-based questions such as: 'What is your speech like, is it better or worse?' and 'Are there other children in your school who

have health problems?' or questions about her emotions such as 'Are you worried or are you fed up?' where Alia hesitated and answered these more monosyllabically. The following excerpt presents Alia's prompt dialogue involving more concrete questions without her parents present:

Dr Louisa Hagan: Have you been to school?

Alia Singh: Yeah.

Dr Louisa Hagan: Every day?

Alia Singh: It was like work experience.

Dr Louisa Hagan: Sorry?

Alia Singh: We had work experience this week.

Dr Louisa Hagan: Okay, so what do you do?

Alia Singh: [cross-talk 00:36:10].

Dr Louisa Hagan: Where did you have work experience?

Alia Singh: Last week.

Dr Louisa Hagan: Last week? (stresses next word) Where?

Alia Singh: In the, in the library.

Dr Louisa Hagan: In the library, okay. Did you like that?

Alia Singh: Yeah.

School and peer relations QoL dimension- Alia responds to the clinician's direct 'minor' questions about school and admits enjoying school work experience

When Alia was not responsive to the prompting by clinicians, the clinicians went further, pressing Alia for her opinions at times, for example:

Dr Louisa Hagan to Alia: Why, why is daddy talking to me? Why don't you talk to me like normal, Alia? (no audible response) (pause) Why don't you talk to me? Why is daddy telling me? Normally you tell me everything.

Non-verbal speech

Consultations with the father present were noticeably different to those when he was absent. The verbal speech was mainly between the clinician and the father and the father interpreted for his wife when he was present in the consultations.

Alia was less verbal, asked fewer questions, was prompted more often and responded *"I don't know"* more. Alia's non-verbal behaviours were noted by the embedded ethnographers 18 times throughout the QoL discussions, in eight of the consultations. The non-verbal contributions included shaking her head, nodding, crying, looking away and avoiding eye contact, looking down, looking sad, sighing, getting out tissues, looking at her parents and smiling. These were non-verbal ways Alia communicated to the clinicians and her parents. The father was present in 16 of the 18 non-verbal references throughout the consultations. Alia's non-verbal communication was usually followed by her father speaking. Often this was on her behalf.

Non-verbal communication tends to be rather minimal in content. When a more expansive response would be appropriate, non-verbal communication by the child opens the way for the parent to expand the child's communication (Clemente, 2009) '*soliciting an answer can be done non-verbally*' (p.880). The following excerpt is an example of how Alia communicated to the clinician non-verbally and how this resulted in the father disclosing further information to the clinician:

Dr Louisa Hagan: I understand, I understand you. She doesn't, she's all right. Okay. How's school, finished? (Alia shakes her head) (no audible response) No?

Clinician asks question to Alia

Alia's non-verbal action signals her father to respond to clinician's request for an expanded response

Hani Singh: I think they have, they have also - have a few weeks more, three weeks more.

Father responds verbally when Alia does not

Alia as Translator

At times Alia's father was unavailable to attend consultations. Alia attended such consultations with her mother. When the father was present he translated into Arabic for the mother and she responded to his speaking in Arabic. In the 8 consultations in which the father was not present, Alia took on the role of translating for her mother.

Alia was relied upon by her mother to translate the clinician's talk but also to translate any questions her mother had for the clinician. At times Alia was translating some difficult discussion and questions between her mother and the clinician, as illustrated in the example below, where the group were discussing the Effects of Tx dimension of QoL:

Alia Singh: My mum is asking: it's not the tumour, yeah?

Child as translator -Alia translated for her mother

Dr Bob Shaw: No, that's not tumour, that's only irritation, that's like inflammation.

Alia Singh: (speaks but not in English for less than 5 seconds).

Alia translates the clinician's answer back to mother

Dr Bob Shaw: Yeah, it's not tumour -

Mona Singh: Okay.

Mother responds to the clinician

Alia's mother spoke in English only occasionally and briefly . She did often appear to speak to Alia in Arabic after the clinician had spoken but before Alia translated for her what the clinician had said:

Mr Kieran Williams: So what are your plans when you go back? What are you going to do? Go to school?

Mona Singh: (speaks but not in English for less than 5 seconds).

Mother speaks

Alia Singh: I have exams.

Alia responds to the clinician after her mother speaks

Mr Kieran Williams: You have exams. When are they?

Alia Singh: August, August.

Responding "I don't know"

Alia responded *"I don't know*" to the clinician's questions nine times throughout the consultations. Alia's father was present eight of the nine times. Answering "*I don't know*" also usually resulted in Alia's father speaking for Alia.

Eight of the nine questions were broad and required a subjective judgement on Alia's part, for example, *"what do you think you want now?"* Alia also seemed to hesitate to answer questions when she was given a choice "*what is your speech like? Better, worse or same?"* If the question was more straightforward, even requiring some judgment such as "are you tired?", Alia managed to answer these types of questions, regardless of who was in the room. Alia was also able to answer in any interaction the concrete questions requiring factual information such as "*do you go to school every day*?".

When asked by her father to tell the clinician about her feelings towards school, she responded "*I don't know*" and her father continued to speak for Alia and advocate her wishes on a subject he knew was of importance to Alia.

However, again on the dimension of school, when the father was absent and unable to support Alia in the consultation, the mother appeared to help Alia in Arabic and Alia was able to answer and present her own priorities regarding school:

Dr Louisa Hagan: Alia, how are you in school, can you follow the learning and teaching and everything?

Alia Singh: (speaks but not in English for less than 5 seconds).

Mona Singh: (speaks but not in English for less than 5 seconds)

Alia Singh: Yeah, I'm fine.

Dr Louisa Hagan: You're fine?

Mona Singh: (speaks but not in English for less than 5 seconds)

Alia Singh: So when I come back, I can't study…like I don't have that much study so, so every Thursday we have a science test…that we took the whole week. So sometimes I can't study like that much after school-

Dr Louisa Hagan: Yeah

Alia Singh: -because I'm tired.

Patient-centred behaviour

Using the perspective of interactional sociolinguistics as described by Tates et al, (2002b) about how '*participants display their orientation to their institutional roles and identities; how they collaboratively co-construct the course of action; and how these discursive constructions structure the ongoing interaction*' (p. 109), the clinician's behaviour emerges essentially as patient-centred behaviour. Such behaviour contributes to the structuring of the interaction and has consequences for the child's participation in the consultation.

In one consultation that both parents attended, Dr Hagan at one point asked the parents to leave the room so that she could speak to Alia alone. From the clinician's statements during the interchange, it was clear that the clinician was interested in eliciting remarks which Alia might not want to make with her parents present. The clinician questioned Alia about further Tx. Between the clinician and Alia, all of Alia's speech was verbal but was in response to the clinician's questions. However, Alia did not ask any questions herself during the dyad interaction, initiated no speech, and initially responded "*I don't know*" when asked to make a major decision without her parents in the interaction (see Section B. below for decision-making). The clinician was seeking something Alia might not say with her parents present, but the result of this one to one conversation was that Alia was less forthcoming than when her parents were present.

In another consultation the consultant pressed Alia for her opinion on her side effects and symptoms:

> *Dr Louisa Hagan: Yeah. Is that difficult? Is sitting on the chair difficult? (pause)*
>
> *Mona Singh: (speaks but not in English for less than 5 seconds).*
>
> *Alia Singh: I'm okay.* } Alia replies briefly
>
> *Hani Singh: She, she, she has, she has [inaudible 00:06:48] but she, she sat, she sat - (speaks but not in English for less than 5 seconds) -*

Mona Singh:(speaks but not in English for less than 5 seconds).

Hani Singh: Yeah, she sat down a few times probably yesterday and today. [cross-talk 00:07:00] -

Father elaborates for Alia

Dr Louisa Hagan to Alia: Why, why is daddy talking to me? Why don't you talk to me like normal, Alia? (no audible response) (pause) Why don't you talk to me? Why is daddy telling me? Normally you tell me everything (pause).

Clinician asks Alia why her father is responding for her

Hani Singh: Yeah, sorry about that.

Dr Louisa Hagan: No, no, no, it's okay, it's okay. No, I just want to know why. Why is that? Is that because you're tired? Is that because you're fed up? (no audible response) Hm? (pause) Does, does your, does your neck hurt more when you lean back like that? Does it hurt more?

The clinician then asks further questions and uses prompts

Alia Singh: Same.

Alia answers when prompted but monosyllabically

Dr Louisa Hagan: Same?

Clinician requests clarification on what 'same' means

Alia Singh: A bit.

Here the clinician addressed a question to Alia and marked Alia's silence, allowing her father a space to respond.

Throughout the consultation, the clinician continued to address her remarks to Alia even though the father might be the only one speaking in response to the clinician.

Parental child-directed behaviour

The parental role holds two aspects in tension. Parents are advocates for and protectors of their children. In these consultations with an adolescent child, the advocacy/supportive

element was observed to emerge at times to support their child's participation in the consultation.

There were multiple times when the father tried to bring Alia into the conversation and engaged in meta-communication, encouraging her to articulate her views. Below is one such example:

Dr Louisa Hagan: How's school, have you done your exams?

Alia Singh: Yeah (quietly).

Dr Louisa Hagan: How did you do?

Alia Singh: (hesitation) Okay (quietly) (pause).

Mona Singh: (speaks, but not in English for less than 5 seconds).

Dr Louisa Hagan: Not happy?

Clinician senses something is wrong and asks Alia if she is not happy

Hani Singh: Yeah. Say what you have to say.

Father encourages Alia to speak for herself

Dr Louisa Hagan: Go on. (pause) You are not happy or parents are not happy?

Clinician presses Alia again to expand

Mona Singh: (speaks, but not in English for less than 5 seconds).

Alia Singh: I am not. (Pause)

Alia answers but leaves a silence after to indicate for someone else to speak

Hani Singh: Erm. She, she, she scored 57 out of hundred. The pass grade is 60… I cannot complain.

Her father on hearing the pause, expands on Alia's answer

In this section of conversation between the three participant groups- clinicians, parents and child- Alia's perspectives and views were recognised and advocated for in the consultation. Together with the clinician, the family developed a picture of how important Alia's studies still were to her. There was a pause after Alia's speech whereby the father allowed time for Alia to continue. When Alia did not continue, her father gave the clinician

details about how much of a tough time Alia had experienced and how it had impacted her school exam results.

Although the father did most of the talking, Alia joined in and interactionally permitted her father to continue for her. From Alia's limited response to her father's encouragement, Alia permitted her father to speak on her behalf. The mother communicated in Arabic throughout the conversation, which added to the co-construction by the family.

The following excerpt shows an instance when Alia did not respond and Dr Hagan advocated for the child regarding Tx and side effects:

> *Hani Singh to Alia: You said to the doctor it depends…if it is damage not the tumour. What do you mean it depends?*
>
> *Dr Louisa Hagan: (pause)…she needs to think about it.*

Even when Alia's views did not reflect her parents' priorities, on multiple occasions the parents still acted as advocates throughout the consultations, as is discussed in the next section.

Parents' and clinician's co-construction of the child's view

Although Alia did not speak at times, her wish to continue to try to balance school with Tx, was advocated by her parents. This was the case even though the parents explicitly stated that Alia's "*health*" (i.e. not school) was their primary concern:

> *Hani Singh: And so she (Alia) did the exam, she did her best, but she, she couldn't make, she couldn't make it, okay. (Alia looks upset). They didn't make up for her also in the, in the summer. Mum is saying that if, if the treatment is going to - (speaks but not in English for less than 5 seconds).*
>
> *Mona Singh: (speaks but not in English for less than 5 seconds).*
>
> *Hani Singh: Can she continue her studies here?...Can she, can she attend a school while she is doing the treatment?*

Father explains Alia's issue with the mother's input

Father translates for mother and advocates for Alia's needs

Dr Louisa Hagan: Yeah, yeah, yeah, yeah, yeah, yeah, absolutely.

Hani Singh: Okay.

Dr Louisa Hagan: Absolutely. There is no point otherwise, okay.

Clinician agrees and also advocates for Alia

The clinician's final remark also took up Alia's concerns. She framed her remarks from the patient's point of view. In Alia's view, the burdens of Tx would be outweighed by allowing her to do what she deeply valued. The clinician brought Alia's view to the conversation, saying that without facilitating school attendance there would be no point in treating. What occurred then is that though Alia remained largely silent, both parents and the clinician continued to discuss the issue from Alia's point of view; thereby bringing Alia's voice into the consultation.

B. ALIA'S PARTICIPATION IN DECISION-MAKING WITHIN THE CONSULTATION

In this section, I turn to participation in decision-making. Coyne et al (2014), in a study of decision-making with children with cancer, made a distinction between major and minor decisions. '*Major medical decisions included investigations, decision to treat, administration of cancer therapies, selecting a treatment protocol, and delivery of medical and nursing procedures. Minor decisions were decisions about how care/procedures would be delivered*.' (p. 275) The distinction made between major and minor decisions in Coyne et al's (2014) study concerned a population different to that of HRBT, with different prognoses and at a different time in the illness trajectory than those faced by Alia and other children in the HRBT project. In this population, both children and parents are involved in major decisions. Nevertheless, the major and minor differentiation is a useful one and is adopted in this Chapter.

Alia was involved in all of the minor *and* more major decisions, which were made jointly with a clinician and/or parent/s. Alia was involved in eight decisions in the consultations. Four of the decisions Alia made were classed as major and four as minor.

During a consultation about further surgery for a recurrence the clinician, at one point, asked parents to leave the room. The clinician proceeded:

Dr Louisa Hagan: Okay. So shall we do the bloods today?

Decision 1- clinician involves Alia in a minor decision

Alia Singh: Okay.

Dr Louisa Hagan: And, and then I'll organise an appointment for you to the - see neurosurgeon and in the meantime, you will think what you want, okay. (pause) What do you think you want now?

Clinician then asks Alia to be involved in a more major decision re: treatment options and next steps

Alia Singh: (pause) I don't know.

Hesitates and answers "I don't know"

Dr Louisa Hagan: Hm?

Clinician prompts for another answer

Alia Singh: I'll do what's needed.

Decision 2- major decision

Dr Louisa Hagan: You'll do what's needed. But, no, you need to do, you need to do what you want to do, not what's need - what's needed.

Alia Singh: [inaudible].

Dr Louisa Hagan: Hm?

Clinician prompts again

Alia Singh: That's what I mean.

Alia reformulates her response from the clinician's response

Dr Louisa Hagan: Okay, okay, shall I call them back in?

Alia Singh: Yeah.

Dr Louisa Hagan: Hm, yes or no?

Alia Singh: Yeah.

Decision 3- minor decision

Dr Louisa Hagan: Is there anything else - this is your chance, tell me, is there anything else you want to tell me that you don't want your parents to know?

Alia Singh: No.

Dr Louisa Hagan: Because I, I, I'm not going to tell them.

Alia Singh: No.

The clinician asked Alia "*what do you think you want now*?" Alia responded "*I don't know*" to the question, referring to the pursuit of further Tx. The clinician's *"Hm?"* presses Alia to expand her statement without suggesting any direction to take in doing so. This indicates that the clinician does not take Alia's response literally, as indicating some sort of lack of essential knowledge. Rather it shows respect for Alia as having her own opinions and only needing the time and support to express them.

Alia then answered she would do what was needed. The clinician then abandoned her neutral stance and challenged Alia's answer. The clinician's challenge opposed Alia's formulation of what was needed to one of what she (Alia) wanted, implying that what Alia perceives as needed reflects considerations other than what Alia herself wants. The clinician thus suggested a particular direction in which Alia should go in reformulating her response. The clinician exhibited a type of patient-oriented behaviour which embodies the principle that patients' decisions should emanate from themselves without external influence.

The obvious external influence in the clinician's view was the parents whom the clinician had asked to leave the room in order to have the conversation with Alia. This is further confirmed by the clinician's remark at the end of the interchange, revealing the clinician's view that Alia's expressions of what she wants would likely be things Alia does not want her parents to know.

Finally, when pressed by the clinician that she should not do what was needed but what she *wanted*, Alia answered again saying that further Tx *was* what she wanted. The clinician was satisfied with this and, with Alia's permission, ended the one to one and called the parents back in.

In the three other major decisions with both her parents in the room, Alia provided an answer quickly and was less hesitant than during the major decision she was involved in without her parents. When asked about what she wanted to do regarding further Tx, Alia consistently presented herself as wanting to try a further Tx. However, she asked to be informed of further information related to the Effects of Tx on two occasions before she confirmed her decision with the clinician. In the third major decision with her parents present, Alia was asked if she would like further oral Tx or did not want any more Tx. Alia

answered ‘it’s okay’. Although the clinician offered Alia the option of stopping Tx, Alia did not take this option:

Dr Louisa Hagan: Okay, okay. Now if it is not, if we say that it is a tumour, then we would need to see what your blood count is like and we would then need to see if there is any other treatment we could give you but I would then want that treatment to be oral, by mouth, so you don't need to have lines and things like that. Would you agree with that or are you at a point where you've had enough of the treatment and you don't want any more treatment because you're fed up. Just tell me.

Clinician asks for Alia’s views on further treatment

Alia Singh: It's okay.

Alia responds briefly

Dr Louisa Hagan: It's okay. Okay. So that is where we are. I think the - do, do you need to go back to UAE soon? Are you under pressure with time?

Hani Singh: I mean our priority is her, is her health now, you know.

Father responds and states the parents’ priority

Thus, the major decisions Alia seemed most unsure about, hesitating in her answers or answering “*I don’t know*”, were during the interaction where her parents were not present. During the instances when her parents were present, Alia made verbal or non-verbal contact with them when faced with a decision.

C. OVERVIEW OF WHICH QOL DIMENSIONS FEATURE MOST PROMINENTLY

Eight of the 11 QoL dimensions appeared in the consultations involving Alia and her family. Control, Disease-related symptoms and Wishes were not present. School and peer relations were the most frequently discussed QoL dimension, with 41 references. The next most frequently discussed QoL dimension was Effects of Tx, which had 15 references, then Construction of normality (13), Construction of the future (8), Family relations (6), Place of care (POC, 5), Time (3), and Holiday (1).

In Chapter 4, School and peer relations ceased being prominent for clinicians and parents at progression; however, in Alia's case, School and peer relations still appeared prominent after multiple progressions for Alia. Alia's parents identified Alia's health as their primary concern, similar to the other parents in the study.

In Chapter 4, the Effects of Tx were seen by parents and clinicians as something the child had to endure to reach the potential of L/T survival. Before progression, other dimensions were not prominent for either parents or clinicians. Clinicians, however, changed their views at progression and other QoL dimensions came to the forefront. In Chapter 5, hesitancy on the part of clinicians to keep treating when the disease had progressed was observed, alongside Effects of Tx and the other QoL dimensions coming to the forefront. Dr Hagan was seen to be repeatedly questioning Alia and her family's choice of continuing further Tx and asking to hear Alia's opinion to ensure it was Alia's choice. The oncology consultant brought up School and peer relations, Effects of Tx, POC and Family relations on multiple occasions during discussions of care and Tx. An oncologist was observed steering the father away from further Tx that would prevent the family from going back to their home country (see excerpt below), indicating that POC was an important dimension to the clinician at that point in the illness:

> *Hani Singh: Is there any experimental treatments that is-be possible for cases like Alia in hospital?*
>
> *Dr Bob Shaw: Yes, but these tumours are very rare, and as I said…you want to go home at some stage. If you go on experimental treatments you will spend months and months again here and I think at the moment we're trying to as well get you home.*

However, POC did not appear to feature in Alia's parents' QoL concerns. Tx continued to be their priority. Chapter 4 presented POC as a varied part of the parents' QoL concept, which was important to some parents and not to others. Alia's parents only talked about POC in relation to going home to UAE to see their family but this was always worked *around* Tx. School and peer relations was the priority over POC for Alia, with School being the driver in the POC decisions:

Dr Louisa Hagan: …if we are not going to do anything between now and 3 months do you want to stay in London?...

Alia Singh: I don't want to [stay in London] because I have school.

However, Alia was seen to consistently choose further Tx when offered, which her family and clinicians understood. She also consistently inquired into the side effects of any Tx discussed, indicating that the Effects of the Tx dimension were of some importance to her. Alia asked multiple times to know about the side effects of medication and Tx she was, or potentially would be, taking. In the example below with the father present, when Dr Hagan was asking Alia for her opinion on entering a clinical trial or not, Alia asked for information about the side effects of the drug before she agreed:

Dr Louisa Hagan: Would you like to go on the study?

Alia Singh: Yeah

Clinician asks question

Dr Louisa Hagan: (no audible response) Hm? (Alia shrugs)

Clinician prompts. Alia responds non-verbally

Dr Louisa Hagan: (pause) Shall I - why don't I find a little bit more about it so I can tell you next time we meet, okay. I can tell you.

Alia Singh: Will you tell me the side effects?

Alia asks for more information on the Effects of Tx dimension as the clinician suggests

Dr Louisa Hagan: Yeah, yeah, yeah, that's what I need to find out so I can tell you side effects, about how much you would need to be the hospital, how the drug given and so on and so forth, okay.

Parents in Chapter 4 continued to normalise the Effects of Tx and accept side effects as part of the course of Tx until the EoL. Alia's parents also continued with their Construction of normality post-Dx and continued to normalise the Effects of Tx after multiple progressions:

Hani Singh: Normal headache…

Similarly, Alia continued with a Construction of normality post Dx that was the same as that noted at time of diagnosis:

Dr Louisa Hagan: When you wake up you have a headache and then the headache goes away? No?

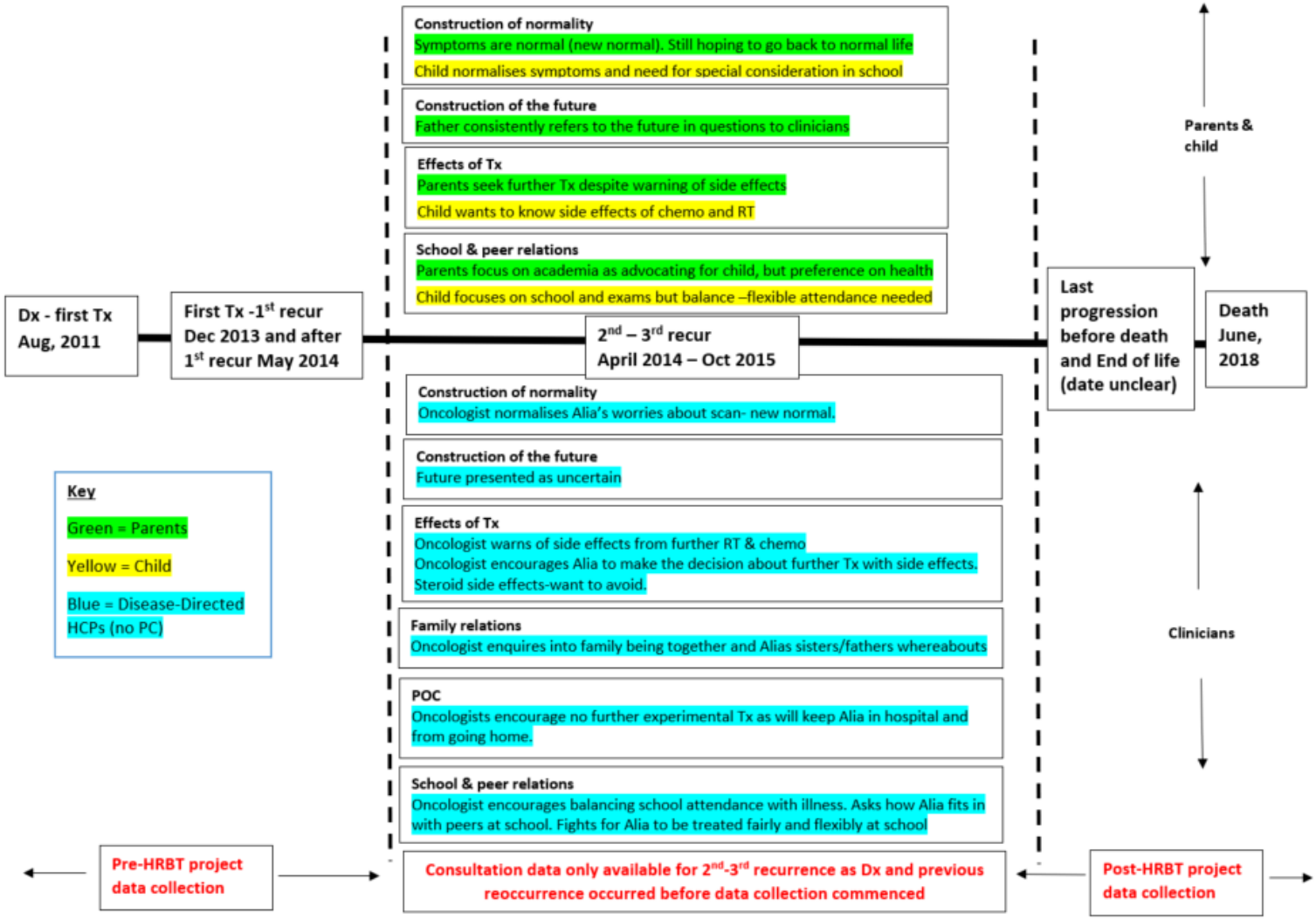
Alia Singh: Like before, the same, normal.

Alia's father repeatedly referred to a Construction of the future in Alia's presence which included hope for L/T survival, as was also reported in Chapter 4. And similarly to the findings in Chapter 4, the clinicians in Alia's case repeatedly responded to manage the parent's expectations about the future by presenting the future as uncertain:

Dr Louisa Hagan: It should at least stop things getting worse…It's difficult to predict.

Because of the continued prominence of the School and peer relations dimension in Alia's case, Family relations did not feature as highly in the QoL talk as other cases presented in Chapter 4. See Figure 15 below for an overview of the QoL dimensions involved in Alia's case over the illness trajectory.

FIGURE 15. QOL DIMENSIONS ARRAYED OVER THE ILLNESS TRAJECTORY – ALIA SINGH



5.3 THE DISCUSSION OF FINDINGS

Chapter 5 set out to add a missing piece to the picture of the QoL of children with HRBTs. That missing piece was the perspective of the children themselves on the QoL. Simplistic, literal approaches to identifying the child's views or voice were found to miss the child's point of view and with it the contribution of their QoL concerns to discussions of their care and Tx. In Chapter 5, an analysis of the consultations of one 14-year-old girl was used to explore these issues about the child's voice.

In this chapter, dimensions of QoL developed earlier were used to identify sections of the consultations for close analysis. The analysis was conducted primarily from an interactionist perspective. The approach broadly fit within what Antaki (2008) identified as discourse analysis. Within that approach concepts and methods derived from interactional sociolinguistics (Clinician and parental roles, patient-directed behaviour) and conversation analysis (Bluebond-Langner et al, 2021) were used.

Finally, through mixed qualitative methods using the dimensions of QoL as topics of discourse (Ochs and Schieffelin, 2011), frequencies of occurrence of the dimensions were taken as indicators of relevance and importance to each of the participants in planning care and Tx. Eight of the 11 QoL dimensions were present in Alia's case. School and peer relations was the QoL dimension discussed most frequently (41 references), followed by Effects of Tx (15 references), the Construction of normality (13 references) and the Construction of the future (eight references). The other QoL dimensions emerged infrequently.

How the findings relate to the participation, QoL and decision-making literature is discussed in the following sections.

A. ADDRESSING THE ISSUES IN THE CHILDREN'S PARTICIPATION LITERATURE

Recognising and addressing the complexity in exploring child participation

Chapter 5 illustrates the complexities involved in children, parents, and clinicians discussing care and Tx together through an in-depth analysis of a single case. The place

of the child in consultations, coupled with often limited verbal contributions, suggests some of the difficulties in 'hearing' the child's voice, and therefore appreciating their perspective. The complexity is because the child's voice is not taking place in isolation; it emerges within a three-way dialogue during a consultation. The participants involved all influence each other and what is said or asked by the child is influenced by the participants in the room. These perspectives and decisions rarely operate in isolation for any patient, even adults (Coulter, 2003). The patient's priorities, wishes and needs emerge in interaction rather than simply being declared or explicitly stated.

Past studies have shown that researchers frequently adopted a dyadic approach to analyse the communication between the doctor and parent and between the doctor and child (Tates and Meeuwesen 2001), even when all were present during the consultation. However, Clemente (2009) argued that to understand the true participation of the child, all three participant groups need to be analysed in relation to one another. Clemente used conversation analysis to interrogate data from video-taped consultations, and identified four practices of child-initiated participation including the solicitation of parental assistance. Other researchers have advocated using observational methods with all three participant groups (Coyne and Gallagher, 2011; Beresford and Sloper, 2003; Weaver et al, 2015).

Chapter 5 suggests and illustrates an interactionist approach which provides further insight into child's participation. The analysis of the data demonstrates how Alia, a child with an HRBT, participated: and how her voice emerged in QoL discussions. Notably, Alia's voice was embedded in interchanges with the clinician and the parents rather than appearing as declamatory statements to an audience of listeners.

The findings from this case support the recommendations from previous studies for observational studies to explore participation. The conversations in most of the consultations in Alia's case were predominantly triadic. The child's voice emerges not only through the child's verbal contributions but from statements and responses of the clinicians and parents throughout the consultations. Measures such as participant's total time speaking fail to reflect the role a participant plays or the impact they may have on a consultation. As Jacoby and Ochs (1995) argued, '*the idea that relying only on the*

informational, semantic, and propositional content of words and utterances will fail to get at what utterances (and silences) might be doing as actions in a sequence of detailed interactional events" (p. 176).

Much of the previous recent research uses interview data and thematic analysis to explore the child's participation in the consultation. In contrast, the analysis presented here demonstrates not only what can be missed from such an approach but also what can be gained by looking interactionally in search of a clearer understanding of participation. Importantly, by looking at the actual on the ground interactions as they unfold over time, insight is gained into the myriad of possible roles children can play in the consultation and the depth and breadth of their participation. Had such an approach not been used, the various ways in which Alia participated would have been missed. Several of the substantive findings here from the analysis emerge only with the use of a triadic analysis/perspective.

Using the in-depth methods of discourse and conversation analysis allowed an analysis that looked past verbalisations and the literal meaning of words. Roles and identities were examined in the triadic interaction, and how adults directed their speech. Taking a discourse analysis approach to statements like "*I don't know*", they are construed as accomplishing actions in conversation rather than as simply reporting on the speaker's mental state of ignorance-or knowledge, and the avoidance of conflict is one such action. In previous research Beach and Metzger (1997) found a number of uses of phrases such as "*I don't know*" including the expression of uncertainty and the avoidance of disagreement or conflict. In this situation, Alia could be doing either in saying "*I don't know",* but this phrase often functioned as an indication for her father to speak for Alia and communicate her views to the clinicians.

Absence of obstacles to Alia's participation

At no point during the consultations were there conversational obstacles to Alia's participation, be it in answering questions directed to her or in her responses. In addition, there was no evidence of attempts to discourage her participation. The clinician

consistently directed her speaking to Alia even when she did not respond verbally. There were no inhibiting reactions when she entered the conversation spontaneously.

Alia herself held many different roles in the consultations, including recipient of information, non-verbal contributions, delegating to an adult, translation, prompted speech, Alia-initiated speech, reformulation, eliciting information (asking questions), and decision-making. There were no interactional or conversational barriers for the child. Alia was able to insert herself into the conversation as she did multiple times throughout the consultations with one or both parents present. This is consistent with the observation that children and young people should have a say in consultations 'as they wish' (Kelly et al, 2017).

The clinicians directed much of their speech to the child whether or not the child was the one responding to the clinician, and during a conversation between a parent and a clinician the child was able to insert herself into the conversation without being invited. And lastly, despite the differences in preference of QoL dimensions, the parents managed to advocate for their child as well as present their own preferences.

Patient and child-centred behaviours of the adults

In addition to observing a lack of barriers to the child's participation, the adults in the interactions displayed child-centred behaviours throughout the consultations. As previous studies have observed (Tates et al, 2002a; 2002b), the clinician exhibited a number of patient-centred behaviours which mitigated against the child being excluded from the conversation. The clinician informed the child, advocated for the child, asked the child direct questions, asked the parents to leave the room, and asked the same question multiple times, in different ways and different interactions with different members of her family present.

The child was embedded in the conversation by the design of the clinician's speech alone. The clinicians addressed the child in their dialogue by directing much of their speech to the child, "*you will feel*" not "*she/he will feel*", even when the child was not verbally responding and her father was the main interlocutor. The father also tried multiple times

to bring Alia into the conversation or to explain her feelings to the clinician, often only speaking to the clinician after he realised Alia wasn't going to speak or continue to speak.

Previous studies have reported that children are often excluded from what becomes a dyadic conversation between the clinician and the parents (Cahill & Papageorgiou, 2007). In the case of Alia, there was clear evidence of parents facilitating rather than inhibiting Alia's participation. There was no evidence that the clinician and parents constructed a conversation from which the child was excluded.

I suggest that the usual reasons given for a child's lack of verbal participation in consultations do not apply to this case, and that some other explanation of Alia's relative silence is needed.

Co-construction of the child's views

The findings presented here are consistent with previous research indicating that relying upon parents is an active choice and a key strategy in child participation (Clemente, 2009; Kelly, et al, 2017). On her own, with the clinician, Alia did not ask questions and answered monosyllabically. Interestingly the child's voice emerged least in dyad interaction without her parents. By removing the parents from the room, the clinician got no closer to the child's perspective than when both parents were present in the consultations. In other interactions where the clinician had isolated the child's voice by asking the parents not to speak, again, Alia responded no differently from when the parents were active in the consultation.

The presence of parents facilitated Alia's decision-making rather than supplanting it. Alia seemed to find it easier to make the major decisions with her parents in the room than on her own. As previous studies have found, Alia's father, like other parents, often allows children to try to respond first before they then respond to the clinician (Stivers, 2001). When he did respond for Alia, it often came after Alia had spoken, with a delay or pause in her speech or when Alia had delegated to her father non-verbally, or verbally in English or Arabic. The interaction between Alia and her father appeared to be bi-directional in that

her father supported his child in the consultation, and Alia often sought her father's support.

Allowing a parent to speak on their behalf and trusting parents to represent a child's view is supported in the literature (Ruhe et al, 2016b; Weaver et al 2015; Kelly et al, 2017). Ruhe et al (2016b) found that some children preferred their parents to be a messenger for information, similarly, Kelly et al (2017) and Weaver et al (2015) found that children trusted their parents (and clinicians) to do what was best for them. Eight of the nine questions that Alia responded '*I don't know*' to were broad, judgement related questions. For these questions, Alia's father would support his daughter in her answer. These findings align with previous findings that when the clinician asks broader questions, the child finds these more burdensome and they are less likely to be answered (Clemente et al, 2012; Clemente et al, 2008). The presence of the parent/s was not invidious but was helpful to the child's participation. The parents understood and acknowledged the difference between their perspectives and that of Alia's, yet still advocated for their daughter's wishes. The parents worked together with the clinician and their daughter to try and balance their needs and co-constructed the child's views and voice.

The child's impact continues even when she is not verbal, for example, when her father asked the clinician if Alia could continue her studies during her Tx. Aware of how important school attendance is to Alia, Dr Hagan responded "*Absolutely. There is no point otherwise".* Another example of this continuing impact is when the father relays to the clinician:

> *Hani Singh: I mean, it's - my view is that her treatment is more important than, than the study, this is my view. But Alia, I know that she's always conscious about study, okay, and the work.*

Alia's parents supported their daughter's wishes and worked with the clinician to find a balance between continuing school in some capacity while pursuing further Tx.

These findings build on prior interactionist research that provides evidence of children and parents working together to jointly construct a child's view (Jenkins, 2015). Jenkins (2015) explored the conversations between parents and their children with L/T health issues about the child's pain. Similar to the present study, Jenkins focused on the parent's

responses and the nature of subsequent talk between the child and parent and found that the family co-constructed the child's views. These findings also align with Clemente's (2009, 2015) work demonstrating that non-verbal behaviours are as important as verbal behaviours in deciphering how a child's views are co-constructed. Parents work together with the child to answer the clinician's questions in the consultation (Clemente, 2009).

The many ways Alia allowed and welcomed her parents to support her or speak for her in the consultations indicated that she relied on both her parents in different ways during the consultation to help co-produce her views.

B. ADDRESSING THE QOL DIMENSIONS OF SCHOOL AND PEER RELATIONS AND EFFECTS OF TX

By combining interactional analysis with the identification of topics of discourse via the dimensions of QoL, differences between child and parent's expression of the QoL dimensions and issues surrounding QOL became boldly apparent. Alia was most involved verbally in the School and peer relations dimension but she asked more questions and initiated more talk on the Effects of Tx. Examination of the School and peer relations QoL dimension revealed 36 verbal contributions by her. The two questions Alia asked about school helped us to understand that school was still important to Alia even at this late stage in the illness, as was having the flexibility to attend school when she felt up to it. Other studies have found that children with cancer who attended school reported feeling better and had an increased sense of independence (af Sandeberg et al, 2010). This differed from the parent's priorities, in which School and peer relations did not feature as highly at this later period in the illness, mirroring findings in Chapter 4.

Such examination also revealed much about Alia's perspectives on the Effects of Tx dimension. Alia made three decisions, asked seven questions, and made two of the three reformulations in the Effects of Tx dimension. The questions Alia asked implied an interest in the side effects of the proposed Tx. Alia used the information she gained about the Effects of Tx in making decisions about her care and Tx. This may reflect a balancing of the burden of Tx versus possible gains related to other QoL dimensions that is different from the balancing by her parents. The parents, similar to the parents in Chapter 4, did

not enquire as frequently about the Effects of Tx. At this stage in the illness, the parents were still willing to trade off QoL dimensions for the pursuit of Tx. However, Alia inquired repeatedly about School and peer relations and the Effects of Tx dimensions of QoL. Previous studies have acknowledged these differences between the parent's and children's judgements of QoL (Eiser and Varni, 2013). Authors conclude that when evaluating HRQoL, both child's and parents' perspectives should be considered for a holistic view and that the differences should be viewed as a source of valuable information rather than measurement error.

The parents and clinicians advocated for Alia's interests too. And interestingly, with the father present, more QoL references were discussed (60 of the 92 total QoL references). It took the input and interpretation of all participants to understand Alia's preferences around the Effects of Tx, School, and what was important to Alia.

C. ADDRESSING THE ISSUES IN THE DECISION-MAKING LITERATURE

Policy guidance and ethicists urge children's involvement and participation in decision-making (Department of Health 2003; Royal College of Paediatrics and Child Health, 2000). Findings from the current study align with Weaver et al (2015), Ruhe et al (2016b) and Kelly et al (2017) that children prefer not to be the sole decision-maker. As these other researchers have found, Alia appeared to be more at ease with making decisions with her parents support than when facing the pressure to make decisions without her parents. Coulter (2003) found that adults had varying participation needs regarding decision-making in the consultation, with many not wanting the burden of having to make Tx decisions.

Studies with children have also stressed the importance of not burdening children with the responsibility of leading on decisions about their care (Deatrick, 1984, Clemente, 2015). Indeed, the interactional analysis provided in this chapter offers insight into what the sharing of a decision entails. By exploring all participants' involvement such as the child permitting a parent to speak on their behalf, or the child just being present and receiving the information from the clinician, a decision has been shared. The child does not have to have verbally affirmed the decision to have shared the decision.

It has been reported that children are involved in the smaller, minor decisions rather than more major decisions (Coyne and Gallagher 2011; Kelly et al, 2017). This is challenged by the current study. Although it is challenging to compare the decisions made by Alia in the HRBT project with the decisions in these previous studies which had greater potential to be life-saving, Alia was involved in many of the minor *and* major decisions regarding care and Tx made jointly with a clinician and/or parent. Alia seemed more comfortable and consistent when making the *major* decisions with both the parents in the room compared to the interaction where she was alone with the clinician in which she regularly paused and answered "*I don't know*".

5.4 CHAPTER CONCLUSION AND TRANSITION TO CHAPTER 6

This chapter set out to explore how children participate in the consultation. Using the close analysis of a single case, the exploration focused in particular on how a child's voice emerged in interactions in which QoL related issues are discussed. With QoL accepted in the literature as a subjective concept, it is important that the child's own views and perspectives are taken into consideration.

Participation took many forms for the child in the case study, illustrating that the total time spent speaking by a participant is an arbitrary measure of the role a participant plays and of the impact they may have on the consultation. A more positive and nuanced portrayal of child participation may unfold when using observational methods of data collection and wider definitions of participation.

There were no obstacles to the child's participation. The clinicians and parents invited the child's participation, listened to her remarks, and put her views forward. The parents should not be seen as taking away the child's voice when the child remains silent. The child's impact continues even when the child is not in the conversation. Parents and clinicians continue to present and advocate the child's views even when their views and priorities may be different to their child's.

The analysis revealed that School and peer relations and the Effects of Tx dimensions were of most importance to Alia. These dimensions were considered in discussions by

parents and clinicians regardless of whether or not the child articulated them herself. The family worked together in the case study to co-produce Alia's perspective with and to the clinician.

In Chapter 6, the results from the previous chapters are brought together and discussed, and the research and clinical implications of the thesis as a whole as well as limitations of the study and opportunities for future research are also presented.

CHAPTER 6 - CONCLUSIONS AND RESEARCH AND CLINICAL IMPLICATIONS

6.0 CHAPTER OVERVIEW

This final chapter concludes the thesis with a summary of the key research findings. This is followed by research implications and recommendations for clinicians and clinical practice. Lastly, the methods of the study are reflected upon and further research opportunities are proposed.

6.1 SUMMARY OF THE THESIS

As work and development of this thesis demonstrates there is a dearth of research on children with BTs at several levels. From understanding children's participation on the ground in consultations, issues of QoL that emerge in the consultation in discussion with parents and clinicians were identified. Parents have different views that change over time, as do the clinicians. The children participate in the interaction with the parents and clinicians and that participation was not hampered by the parents or clinicians.

Chapter 1 presented the research problem and identified the gap in the literature that the thesis addresses. The increase in survival in cancer led to a focus on survivors of cancer and the quality of that survival or 'quality of life' in survival (Spieth and Harris, 1996). Further relevant developments included the call for adding assessment of QoL in clinical trials of cancer interventions for children and young people. QoL in both contexts has been assessed through a variety of measures. The proliferation of such measures has been worrisome to some important researchers in the field (Eiser and Morse, 2001a; Wallander, 2001; Haase et al, 1999; Anthony et al, 2017), suggesting a focus on the measurement itself while neglecting to reflect upon what was actually being measured.

The focus on measurement leads to a neglect of what QoL is in the "minds" of those for whom it is of the most immediate importance: the children themselves, their parents and the clinicians who care for them. QoL is very much a subjective concept, yet gaps remain in the understanding of what QoL means from the perspective of the child, parent and clinician. Including the child's voice in discussions about their own care and Tx has also been recently identified as important (House of Commons Health Committee, 1997; Royal

College of Paediatrics and Child Health, 2000; Department of Health, 2003). Thus, this thesis aimed to understand the perspectives of the parents and clinicians with a particular focus on capturing the child's voice in the consultation and at a unique and difficult time for families, during Tx decisions when cure is not the goal.

Chapter 2 situated the project in the relevant literatures and presented my systematic review (Beecham et al, 2019). The review recommended the exploration of QoL concepts of children who are undergoing active Tx. Hinds (2010) also stressed that QoL can vary as severity of Tx changes and continues to do after Tx ends, hence the need for investigation of QoL over time. The chapter discussed how there are still many gaps regarding the understanding of the subjective and dynamic concept of QoL and the paucity of research on involving the child in the clinical consultation, what children's participation involves, or what form it should take.

Chapter 3 presented the theoretical perspective and methodological approach which underpins this thesis. The chapter gave an overview of the HRBT project that provided the data for this thesis. Chapter 3 described how an interactionist perspective provides the theoretical foundation of the project and influenced every stage of this mixed qualitative project. Finally, the thematic and constant comparison analysis methods used in Chapter 4, and the discourse and conversation analysis used in Chapter 5, were discussed.

Chapter 4 explored how the QoL of children with HRBTs entered into the consultation for parents and clinicians from the HRBT project, using a set of 'dimensions of QoL' developed in this thesis. The chapter presented the dimensions of QoL, organised under four domains, highlighting the differences in key dimensions between the two participant groups; parents and clinicians. The findings were then discussed in relation to previous literature in the area, including issues in the QoL measurement literature. It contributes to filling a gap in the literature that to date has focused on quantitative measuring of QoL and illustrated that individuals' QoL concerns are dynamic and subjective.

The chapter provided a qualitative, unique and detailed description of the QoL-related issues that arose in consultations for the parents and clinicians. The thematic analysis and constant comparison found that the relevant concerns varied between the two

participant groups and changed for each group over the course of the disease. For example, School was prominent for both clinicians and parents until the progression of the disease, and then Family relations supplanted it in prominence for these two groups. Some domains of QoL, Normality and Future, also changed in content over the course of the illness. Early on the future for the parents contained hope for survival and attainment of a cure. The progression of the disease led to their holding multiple views of the child's future and then only at the EoL did the future become short and the parents' focus shifted to other QoL dimensions. For clinicians, the future became shorter earlier on, at progression. Normality and Future playing large roles in the QoL concepts of parents and clinicians supports previous research that interpreted QoL as a new normal (Clarke-Steffen, 1997; Deatrick et al, 1999; Van Schoors et al, 2018; Beecham et al, 2019). The thesis also builds on the theory of redefinition of normal from Bluebond-Langner (1996) where parents keep '*expanding the realm of normal'* (p.168) to accommodate the child's condition and Tx. Bluebond-Langner found that families kept redefining normal until they could no longer contain the intrusion of the illness. The current thesis adds to these previous studies by demonstrating how Constructions of normality and the future change for parents and clinicians in different ways and over time for children with HRBTs.

QoL dimensions could appear differently in the discussions and decisions for each participant group. For example, the QoL dimension Place of care (POC) varied in significance even within the parent participant group. For some parents, it was important to include in decisions about care and Tx, while for others it did not appear in their discussions.

Appreciating the variability and contextual nature of these dimensions contributes to the literature which aims to understand the QoL perspectives of the participant groups involved in the care of children with HRBTs. The findings have cautionary implications for the cross-sectional use of QoL measures. For example, in current paediatric neuro-oncology clinical trials, the administering of the PedsQL measure is required as part of current trials. Trials take place over an often long period of time which may involve relapses of the BTs. This thesis shows that QoL assessment may change over time, indeed what QoL aspects are important may change over time and be unrelated to factors of the drug/intervention itself. These changes need to be integrated and interpreted in

light of the findings of this thesis as the changes may introduce confounding factors to the assessment of QoL in the trial.

The results of this thesis also help to guide clinical practices with regard to exploring QoL perspectives in the consultation, as explained in the clinical implications in section 6.3.

The child's participation and involvement in the QoL discussions were potential topics for the analysis for Chapter 4. But, because of the children's limited verbalisations, Chapter 4 essentially presented the parents' and clinicians' perspectives only. The children's findings were found to be too limited to extract patterns or trends from them. Children were approached as little adults, a trap researchers regularly fall into. However, this led the thesis in a direction to better understand the child's views by using different methods to unpick QoL issues for the child and to see how these are manifested in and impact the consultation. As well as gaining an understanding of a child's QoL concept, Chapter 5 also filled gaps in the current evidence base on how to explore the child's voice and understand how children participate in the consultation. Two recent reviews of the literature have called for further research to explore children's participation in consultations and decision-making (Coyne, 2008; Davies and Randall, 2015).

Chapter 5 focused on a single complex case to explore a different approach with which to identify QoL-related issues which were important to the child and to consider how a child might bring these into the consultation in a way other than explicit, direct verbalisation. This was an alternative route to finding the child's voice; indeed, it was an alternative understanding of having a "voice" in social interaction. This meant learning from previous studies who recommended taking a wider approach to participation that included non-verbal as well as verbal speech, using discourse and conversation analysis (Clemente, 2015). Also learning from previous literature that all participants in the consultation needed to be included in the analyses, focusing on triads rather than dyads (Tates and Meeuwensen, 2001; Gabe et al, 2004). Using a wide view of participation and approaching the consultation as a triadic process, the findings revealed the impact a child has even when he/she is not the speaker in the conversation. The importance of parents and clinicians engaging in child-centred behaviours, presenting and advocating the child's views even when the child did not speak, was noted.

The findings from Chapter 5 suggest that QoL for children with HRBTs is centred around School and Effects of Tx. Alia contributed verbally the most in the School and peer relations dimension but she asked more questions and initiated more talk on the Effects of Tx. Previous studies found that school remains an important part of life for children with cancer (Rindstedt, 2015; Vindrola-Padros, 2012). Rindstedt (2015) reported school to be a recurrent topic in the narratives of two children with leukemia. Alia appeared anxious about continuing with her studies at school throughout the study. Vindrola-Padros (2012) also found that the interruption of school was particularly difficult for children with cancer. The lead clinician in Alias care attended to Alia's need to attend school flexibly, however a recent study found that clinicians need to improve the provision regarding education for children that have undergone cancer Tx (Thornton et al, 2022).

Previous studies have also found Tx burden or symptom distress to be related to QoL (Rosenberg et al, 2016; Zeltzer et al, 2009). Rosenberg et al (2016) explored children with cancer including BTs and found 11 distressing symptoms to be related to decreased QoL. Zeltzer and colleagues found poorer physical QoL domains were reported particularly in the children with BTs. Like much of the QoL evidence base however, both these previous studies were based on quantitative measurement and Zeltzer et al (2009) used a survivor population. In a recent study Requena and colleagues (2022) found that children as well as parents and clinicians normalised the symptoms experienced during cancer Tx which led to the inadequate management of symptoms. Alia's recurrent initiation of talk around Effects of Tx challenges this finding. However, authors acknowledged the underrepresentation of children and particularly younger children in their sample and recommend further research in this group too.

This further understanding of child participation of children with HRBTs, adds, from a different methodological perspective, to the current literature and recommendations by other studies that recommend a wide and flexible approach to participation (Clemente, 2009; Kelly et al, 2016; Stivers, 2001). The findings also challenge previous studies which stress the need to isolate the child's voice to understand participation or focus solely on the verbal contributions of the child (Olechnowicz et al, 2002; Miller and Harris, 2012).

This thesis also explored participation in a unique paediatric palliative population where consultations can be highly stressful and time-sensitive decisions are required. In so doing, the findings of this thesis can be of use in communication training -particularly in the conduct of the clinical consultation (see section 6.3, of this chapter).

6.2 RESEARCH IMPLICATIONS

In this section, the research implications of the methodology used are outlined for QoL research and the implications of the findings related to parents' and clinicians' perspectives on QoL. Finally, the research implications are presented relating to the child's participation in the consultation and the child's QoL perspective.

6.2a The method- prospective ethnographic longitudinal study

This thesis used data from the HRBT project, a prospective, ethnographic, longitudinal study. The insights learned from using a study conducted in real-time highlight the benefits of this prospective method of study. Using an interactionist perspective, all groups in the interaction were observed and examined together. Changes were observed in the dimensions of QoL for the participant groups. This method allowed views to be observed as they were discussed in the interactions and over time. These changes would have been missed had data been used from a single point in time. It was the continuous nature of data collection over 20 months that enabled change to be observed. This avoided reliance on people's memories as well as on their reconstructions of past events as would be the case if retrospective interview data had been used. To provide optimal recommendations for clinical practice, research should involve clinical practice observed in real-time as the interactions play out and as consultations are conducted.

Researchers going forward need to consider conducting ethnographic prospective studies to understand phenomena as they occur. Researchers have the pressure of regularly publishing papers for peer-review journals, which is difficult to manage during longitudinal ethnographic studies. More generally, funding bodies, ethics committees and clinicians themselves may be reluctant to support or conduct participant observation

studies. Ethics committees need to appreciate the value that these studies provide and help to facilitate these studies. Ethics committees often focus more on what they perceive as a burden on participants and therefore provide further obstacles to these types of studies. Previous research has shown that participants did not find the studies burdensome (Olcese and Mack, 2012).

It is important that funders understand the difference between ethnographic designs and quantitative designs. Ethnographical on the ground studies, are time-consuming and therefore more expensive than interviews or surveys, but the insights they provide cannot be replicated via any other method (Dixon-Woods, 2003); especially in healthcare, when there are multiple clinicians, specialties, teams, consultations and encounters in which the families are involved.

6.2b Research into QoL

The QoL dimensions developed in this thesis have implications for the field of QoL as it continues to be studied. As acknowledged and discussed in the systematic review (Beecham et al, 2019), quantifiable measures of QoL in health services, government policy and funding as well as in clinical trials and research have an important place in an ever resource-limited national health service. However, the findings in this thesis raise the question of whether currently available measures of QoL are consistent with the understandings of QoL found in the study and more significantly with the understanding of similarly situated patients, parents and clinicians.

The dynamic, subjective and changeable dimensions of QoL discussed in the thesis query the validity of current static, generic tools to measure QoL:

> '*Validity assesses whether the instrument measures what you intend to measure, e.g. QoL. There are several questions to be considered when assessing the validity of measures: Does the measure appear to cover aspects that are relevant to the areas of enquiry of the study, as perceived by patients, their families and relevant users (face validity)*' (Higginson and Harding, 2007, p. 100).

QoL perceptions changed for each participant group over time and in differing ways. Past studies have also found a that one size fits all approach to the subjective phenomenon of QoL is a limited approach to understanding QoL (Haase et al, 1999; Anthony et al, 2017; Eiser and Morse, 2001b). This thesis supports this view and argues for a subjective and dynamic approach to understanding QoL. Researchers are encouraged to use the findings in this thesis to contribute to the refinement of measures used in all areas of QoL research, trials and policy. For example, as mentioned in the previous section, in clinical trials, rather than a one size fits all tool used at one time point in the trial, a more subjective tool that reveals an individual child's needs should be administered at multiple times throughout the trial that will reflect the changes in QoL.

6.2c Research into child participation

By reducing participation to counting the number of verbal utterances alone, previous studies miss much of the child's involvement in clinical consultations. Employing such an approach in this thesis could have led to the conclusion that children had very limited expressed views on QoL. The child's voice in discussions of QoL was explored in this thesis through a wide and nuanced triadic approach to participation. A child participation codebook or 'framework' (see Table 4. Chapter 5) was developed through the literature and the data as a way to interrogate the data and understand child participation.

The framework took the non-verbal and wide-ranging types of participation that had previously been found in the literature and added further types of participation found within the data, to provide an in-depth, more fully realised understanding of child participation. This framework resulted in a comprehensive approach to the many ways a child can participate in the consultation.

Researchers would benefit from exploring participation using this expanded and more nuanced approach. Similarly, so would policymakers, as it would lead to recommendations for involving the child that take into account the family as a whole, not just the child alone. Understanding that the child can participate in multiple ways and when they want helps us to appreciate that when a child does not speak, it is not

necessarily because they are prevented from doing so, it might be because they choose not to. Not speaking can be as much a demonstration of child agency as speaking.

6.3 CLINICAL IMPLICATIONS

In this section, the clinical implications to be taken are presented from Chapter 5, the child's participation in consultations featuring QoL concerns, for clinical implications on Chapter 4 see the Discussion section of Chapter 4.

6.3a Clinical implications from the parents' and clinicians' perspectives on QoL

See Chapter 4 Discussion section.

6.3b Clinical implications from child's voice in QoL concerns in the consultations

Child participation in the clinical consultation

The findings in Chapter 5 on the child's voice in QoL discussions have implications for practice. The findings suggest there are ways that clinicians can promote child participation in clinical consultations. Communication training could incorporate findings on the child's participation found in the literature and further supported by the case study in this thesis as a first step in widening both the meaning and approach taken to what constitutes child participation and how it is best facilitated and realised. Including the child in the consultation does not always have to result in a verbal response from the child for meaningful participation to have taken place. Despite receiving most verbal responses from the parents in the interaction, clinicians can be encouraged to direct their speech to the child (if they do not already), unless they are, for example, specifically asking a question or replying to a parent. The child will then have opportunities to contribute if he/she chooses to.

This thesis supports previous studies that non-verbal behaviour is especially important to observe. If the child is creating space in the conversation which the parent naturally fills, the parent need not be seen as excluding the child from the consultation. Such behaviour

can reflect a constructive relationship between the child and parent in which the child chooses to rely upon the parent to represent them. Therefore, training is needed to help the clinician pick up on subtle non-verbal behaviours the child uses to signal to the parents or clinicians that they have something to contribute to the interaction or they wish for support in the interaction.

Further, teaching clinicians how to navigate the triadic interaction could help clinicians to manage the parents' often different perspectives in a way that improves overall outcomes. Drawing on existing examples, here and elsewhere, instruction can include techniques to help family members to be facilitators of child participation as well as techniques for identifying when they are acting as a possible obstacle. As evidenced in Chapter 5, the parents were found to help co-construct the child's views, as can the clinician, and can provide useful insights into the child's feelings.

Communication training will present scenarios for the clinician to practice where clinicians can work through conversations with the child alone and with parents present to understand which situation the child is likely to be more open in. Facilitators will highlight that it does not change the child's definition of the social situation or setting when that is what inhibits the child from speaking *'freely'* about certain topics (Cf. Bluebond-Langner, 1978, p.10).

Child's perspective on QoL concerns

School and family remained a constant priority for Alia. This aligns with previous research findings (Glasson, 1995). Hinds et al (2004) also found that more generally, QoL domains for children did not change over time.

Our finding of co-construction of QoL perspectives perhaps parallels the use of proxies alongside child reports. The finding supports previous research that advises using the parents' views and other significant individuals in the child's life to get a full and holistic picture of the child's QoL concerns (Eiser and Morse, 2001a).

6.4 REFLECTION ON METHODS USED

As mentioned in section 6.2a, there are many strengths to using a prospective, longitudinal ethnographic design, including the ability to follow change over time, allowing for the capture of actual experiences of children in real-time as the discussions and decisions unfold and the elimination of such issues as recall bias and interpretation of the question. However, the nature of an in-depth ethnographic study means the number of participants that can be followed is limited. Nineteen families were followed in the HRBT project which, although small, is roughly 10% of the total number of newly diagnosed HRBT patients in the UK during the 20-month period. In this respect, the sample is a significant portion of the UK population of such children.

The HRBT project was a single-institution study. Ideally, it was hoped that as a tertiary referral centre it would have received a relatively balanced segment of the overall HRBT population. As it transpired, the tumour types were representative but the population may have been skewed in other ways. The tertiary centre takes on very young cases which will have reduced the study's median age (seven years old), below the median age for this population in the UK (five years old). Whether this would have resulted in having more robust data from children suitable for analysis in Chapter 4 is unknown. Chapter 4 was only able to include the comprehensive views of the parents and clinicians due to a lack of verbal contributions in the consultations. However, this is a common difficulty in paediatric palliative care with this population.

Constant comparison was used as a principal method of analysis. A sufficiently broad selection of cases was recruited that would have included counterexamples, negative findings, and challenge cases. The HRBT project aimed to include every eligible case presenting to the study paediatric hospital during the 20-month period of study. However, some cases were not recruited. This was because of resource limitations, i.e., the number of ethnographers available.

The sample was ethnically, religiously, and socioeconomically diverse. Most previous research in paediatric palliative care lacks the engagement and inclusion of fathers (Nicholas et al, 2020, Bluebond-Langner et al, 2013b). The HRBT project was unique in that there were almost equal mothers and fathers taking part (17 mothers, 15 fathers).

However, the aim of qualitative research is not merely to generate statistically significant results but to uncover process, experiential phenomena, and to understand the 'what, how and why' (Giacomini & Cook, 2000) and search for patterns. Such a view on the relation between qualitative and quantitative research is also stressed by Long and Marsland (2011) who underscore '*the utility of combining the process-oriented, explanatory potential of qualitative approaches with the statistical benefits of quantitative methods'* (p.84) and by implication identify the unique contribution of qualitative research.

Even for articulate children, speaking took up a relatively small portion of the consultation. This led to a consideration of how participation in consultation could be best explored and presented. The decision was taken that the case illustrating the greatest number and forms of participation would allow for the most fruitful exploration. The goal was to identify processes within the consultation and put forward hypotheses of what occurs between parents, children and clinicians. Whether the conclusions of Chapter 5 are correct can only be confirmed through further similar studies on different samples. The approach succeeded in extricating the QoL issues for the one child, but the search for variations and challenging cases needs to be pursued.

Using an interactionist perspective and approach throughout the thesis has led to a detailed and holistic description of QoL for each of the participant groups. This perspective encouraged the data to be interrogated in such a way that subtleties in the interactions emerged and recommendations could be written that are representative of real-life consultations.

The HRBT project had limited researcher bias. Two ethnographers conducted the majority of the observation, and because of the length of time they were in the field, their presence would have soon become '*unremarkable*' (Bluebond-Langner et al, 2017).

The HRBT project set out to explore decision-making and all aspects of communication, and while study of QoL was not explicitly in the original set of aims, QoL can be seen as a part of communication and decision-making. Also, as is usually more common in *quantitative* research, large databases are often used for multiple purposes such as to develop new hypotheses and test hypotheses and research questions. In qualitative research, the relation between an original research question and the data is different to

that in quantitative research. In qualitative research, the research question may evolve over the process of analysis. As such, it is arbitrary to exclude posing additional questions to a data set. The HRBT data set is a large *qualitative* data set comprising thousands of transcripts of consultations and other encounters. QoL too, as it was approached in this thesis, was explored widely through dimensions devised through the literature and data and therefore the excerpts taken from the data set were rich and full.

The decision was taken to only include the verbatim transcripts with added fieldnotes as this fits the problem of the thesis. I used an interactionist perspective to understand these encounters. There are other parts to the data set (such as informal encounters and interviews) but they did not lend themselves to the analysis that was required for this thesis. This thesis explored a new and developing area of research, using the actual on the ground interactions of consultations of children with HRBTs where cure was not the goal of Tx and with particular attention to issues and concerns of QoL. There are other related issues that could be explored using this database such as the patient/parent/clinician experience and their reflections on it but this was not the focus and is beyond the scope of this thesis.

Reflecting on my experience using transcripts from the HRBT project and multiple qualitative methods, I come away with the following reflections. I brought my experience in the area of qualitative methods and paediatric PC to the analysis of the HRBT project data which enabled me to have a grounding in terminology used by the PC team for example and some knowledge of key theories and issues in the area. But I also brought almost complete naivety (I only observed ~ six encounters in the HRBT project) of the HRBT project data collection and of cases/families and disease-directed clinicians on the project. I think approaching the data with minimal expectations or experiences of the families themselves would have helped to decrease any biases that may have formed had I been one of the main ethnographers on the project.

Despite my experience with qualitative data, I had never experienced the volume of data that came from the HRBT project. I was quite overwhelmed with the amount of data at first but soon made a plan to handle the volume of data in NVivo. Clearly labelled and dated files were enormously helpful with juggling the sometimes concurrent analyses I

was conducting for different parts of the PhD (and the wider HRBT project). Being involved in the wider HRBT project and analysis for other papers (see Appendix 8), provided me with a team of other researchers that PhD students would otherwise not have access to. This helped not only with the isolated feeling that PhD students can experience but practically, with the checking of information such as gaps in any information on encounters or helping with understanding the make-up of cases/families on the project.

If the PhD study was to be conducted again, in retrospect, I would make the following change. The original intention was to compare all three perspectives and I approached the data of parents and clinicians the same as I did the children. Essentially I treated the children as 'little adults' which was an error on my part. In retrospect, I would have been more child-centred and used a different and a wider, more nuanced approach to understanding the child's QoL perspectives, staying within the interactionist perspective, from the start of the study. This would allow me to explore more children's views than the one case I was able to explore in the thesis.

6.5 RECOMMENDATIONS FOR FUTURE RESEARCH

It would be useful to speak to the three participant groups and ask generally about the findings from this study to provide further support for (or question) the QoL dimensions developed. Families in a similar situation with a child with a BT or another life-limiting/threatening illness could be interviewed about the QoL dimensions developed in the thesis to understand if the dimensions are relevant to wider populations.

Learning from the limited data from children in Chapter 4 and the case study results in Chapter 5, future research into children's views of QoL could stratify by age to recruit more children of verbal age who would participate more in the consultation. This would allow for further understanding of the child's QoL issues and concerns and would help to support or challenge findings from the case study on participation. Using a multi-site study would circumvent the issues of small sample sizes due to the rare nature of these BTs.

This thesis has highlighted the benefit of conducting research in real-time, in vivo, as the events unfold. Further research could enquire into the results of other methods used in the HRBT project. The original HRBT project utilised interviews in addition to participant observation. Only ethnographic data was used in this thesis, but a future study could compare the interview data with the ethnographic data to understand the difference between these methods. Interviews have been critiqued as they rely upon recall and therefore may present a view reflecting the subject's present state as well as the past experiences which are the object of the interview. Comparing the actual events occurring in the consultations with what the parents and clinicians portrayed in the interviews would provide evidence of the different information each of these methods provides. It would also provide further evidence on the benefit of participant observation which is often overlooked for the quicker method of interviews.

A study exploring QoL in children with low-risk BTs would help to understand the phenomenon in children who will survive. It would be important to understand if the QoL dimensions were similar to those present in children with HRBTs or if, as found in the systematic review of studies of BT survivors, the dimensions were different (Beecham et al, 2019).

Lastly, as outlined in the clinical implications section, a future study could explore implementing some of these clinical recommendations into training courses in medical schools or for continuing, advanced communication training. It has been repeatedly reported that communication training for clinicians in palliative care is lacking (Baker et al, 2007). Social science research on communication is not reaching physicians or clinical practice (Gulbrandsen et al, 2022). Gulbrandsen and colleagues highlight the importance of understanding the "physiology of interaction" from social scientists to improve clinicians' communication skills. Only experienced clinicians are often trusted with the difficult conversations with children and younger less experienced clinicians are often not in the room. Training in what is important to the families and how to get the best out of children in a triadic interaction could become part of a training course or continued professional development that clinicians are required to undertake.

6.6 CLOSING SUMMARY

This thesis explored children's, parents' and clinicians' perspectives on QoL-related issues in consultations about the care and Tx of children with HRBTs. The current QoL literature focuses heavily on the quantitative measuring of QoL and fails to understand the subjective experiences of the participants involved in the child's care and Tx. This thesis helped to fill this gap. Using an interactionist perspective and data from a prospective, longitudinal, participant observation study, this thesis provides a rich description of the children's, parents', and clinicians' perspectives on QoL-related issues as they emerge in crucial conversations about children with HRBTs.

The thesis developed dimensions of QoL to understand the participant groups' perspectives. The participant groups were seen to hold differing perspectives on QoL that changed and adapted to circumstances at different times in the illness. The dimensions changed in prominence and in nature, suggesting that QoL is not only multi-dimensional but multi-faceted, posing further challenges for the current static QoL measures in use.

The limited data on the child's perspective from verbal contributions alone in Chapter 4 directed the thesis towards a wider and more inclusive approach to understanding the child's view, another area of limited exploration in the literature. A more positive and nuanced portrayal of child participation resulted from the interactionist and triadic approach in Chapter 5 of the thesis. Chapter 5 revealed that the child participated in a myriad of ways in the consultations in which QoL was discussed. A child's impact in the consultation was observed even when the child was not the main speaker in an interaction. The parents, clinicians, and child together co-constructed the child's view in discussions of QoL. This gives insight into what the *sharing* of a decision truly means.

The insights identified in this study provide further support for conducting prospective longitudinal participant observation studies. The changeable QoL dimensions and triadic approach to interaction provide a basis for the design of education and training which can support clinicians in these complex and emotive consultations. These findings also invite policymakers and researchers to reflect upon the QoL measures which they employ to design interventions and to provide services to ensure that they reflect the uniquely

important perceptions of the individuals comprising the populations whom they aim to benefit.

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APPENDICIES

APPENDIX 1 – THESIS DISSEMINATION

Work stemming from the thesis has been presented in 10 international and national conferences including an invited plenary talk in Rome and has contributed to two published peer-reviewed articles:

- **Beecham, E**., Langner, R., Hargrave, D., & Bluebond-Langner, M. Accompanying children - listening to their concerns and fears of living: Quality of life. 5th Maruzza International Congress on Paediatric Palliative Care, 25th-28th May 2022. (Oral presentation).
- **Beecham, E**., Langner, R., Hargrave, D., & Bluebond-Langner, M. Children's and young people's participation in consultations with paediatric oncologists: An analysis of on the ground interactions. In 12th EAPC World Research Congress Online, 18th-20th May 2022. (Oral presentation).
- Bluebond-Langner, M., Hall, N., Vincent, K., Henderson, E. M., Russell, J., **Beecham, E**., Bryan, G., Gains, J. E., Gaze, M. N., Slater, O., Langner, R. W. & Hargrave, D. (2021). Parents' responses to prognostic disclosure at diagnosis of a child with a high-risk brain tumor: Analysis of clinician-parent interactions and implications for clinical practice. *Pediatric Blood & Cancer*, 68(3), e28802. https://doi.org/10.1002/pbc.28802. (See Appendix 8 for full version of paper).
- **Beecham, E**., Langner, R., Hargrave, D., & Bluebond-Langner, M. Parents and clinicians' reconceptualisation of the future for children with high-risk brain tumours as revealed in consultations and home visits. In 17th World Congress of the European Association for Palliative Care. Online, 6th - 8th October 2021 (Poster presentation).
- **Beecham, E**., Langner, R., Hargrave, D., & Bluebond-Langner, M. Exploration of the concept of 'normality' in interactions between clinicians, parents and children with a high-risk brain tumour. Marie Curie Palliative Care Research Virtual Conference 2020, 5th November 2020 (Oral presentation).
- **Beecham, E**., Langner, R., Hargrave, D., & Bluebond-Langner, M. Children's, parents' and clinicians' perspectives of 'normality' as revealed in conversations between clinicians, parents and children with a high-risk brain tumour: Implications for practice.

In 11th EAPC World Research Congress Online, 7th - 9th October 2020 (Oral presentation).

- **Beecham, E**., Langner, R., Hargrave, D. & Bluebond-Langner, M. (2019). Children's and Parents' Conceptualization of Quality of Life in Children With Brain Tumors: A Meta-Ethnographic Exploration. *Qualitative Health Research*, 29(1), 55–68. https://doi.org/10.1177/1049732318786484. (See Appendix 3 for full version of paper).
- **Beecham, E**., Langner, R., Hargrave, D. & Bluebond-Langner, M. Quality of life in children with high-risk brain tumours: children's, parents' and healthcare professionals' perspectives over the course of the illness. Association for Paediatric Palliative Medicine Research Day, November 2019 (Oral presentation/workshop).
- **Beecham E**. Langner R. Hargrave D. Bluebond-Langner M. Exploration of the Term Quality of Life' in Consultations of Children with a High-Risk Brain Tumour over the Course of the Illness. 16th World Congress of the EAPC, Berlin, May 2019 (Oral presentation).
- **Beecham, E**., Langner, R., Hargrave, D. & Bluebond-Langner, M. Exploration of the term quality of life' in consultations of children with high-risk brain tumours over the course of the illness. UCL Cancer Domain Symposium, May 2019 (Oral presentation).
- **Beecham, E**., Langner, R., Hargrave, D. & Bluebond-Langner, M. Children's and Parents' Conceptualisation of Quality of Life in Children With Brain Tumours: A Meta-Ethnographic Exploration. ICH, UCL, November 2018 (Poster presentation).
- **Beecham, E**., Langner, R., Hargrave, D. & Bluebond-Langner, M. Plenary panel: Controversies in practice & policy in quality of life research: What does the evidence tell us? What is needed in way of further research? Exploration of the term quality of life' in consultations of children with a high-risk brain tumour over the course of the illness. 8th International Cardiff Conference on Paediatric Palliative Care. July 2017 (Oral presentation).

APPENDIX 2 - GLOSSARY OF TERMS USED IN THE THESIS

1. Children

The term children is used to refer to all patients in the study aged under 18.

2. Consultation

The term consultation in the thesis refers to the clinical meeting between a clinician and the family. This can take place in the hospital or home.

3. Interaction

An interaction refers to the communication act between the participant groups within a consultation.

4. Disease-Directed/ Non-Disease-directed

Disease-directed refers to therapies or interventions aimed at stopping or slowing the progression of the disease.

Non-disease-directed refers to therapies or interventions directed at adverse effects of treatment, sequelae of disease (e.g., seizures), discomfort, or pain—what we and others call symptom-directed or palliative care.

5. Healthcare professionals/ Clinicians

I refer throughout the thesis to healthcare professionals (HCP) and clinicians interchangeably and both these terms refer to doctors or clinical nurse specialists.

6. Triad

Triad in this thesis refers to three or more participants in the group.

7. Recurrence/progression

Cancer that has recurred (come back), usually after a period of time during which the cancer could not be detected. The cancer may come back to the same place as the original (primary) tumour or to another place in the body.

8. Meta-communication

The term meta-communication refers to talking about talking or communication about communication. For example, when a parent encourages their child to speak for themselves, this is meta-communication.

APPENDIX 3- SYSTEMATIC REVIEW

Research Article

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Children's and Parents' Conceptualization of Quality of Life in Children With Brain Tumors: A Meta-Ethnographic Exploration

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Abstract

The concept of quality of life (QoL) is used in consultations to plan the care and treatment of children and young people (CYP) with brain tumors (BTs). The way in which CYP, their parents, and their health care professionals (HCP) each understand the term has not been adequately investigated. This study aimed to review the current qualitative research on CYP, parents' and clinicians' concepts of QoL for CYP with BTs using meta-ethnography. Six studies were found, which reflected on the concept of QoL in CYP with BTs; all explored the CYP's perspective and one study also touched upon parents' concept. A conceptual model is presented. Normalcy (a "new normal") was found to be the key element in the concept. This study calls for a conception of QoL, which foregrounds normalcy over the more common health-related quality of life (HRQoL) and the need to understand the concept from all perspectives and accommodate change over time.

Keywords

survivorship; adaptation, coping, enduring; cancer; psychosocial aspects; illness and disease; children; resilience, resistance; lived experience; health; experiences; quality of life, qualitative. Geographic: Taiwan; Method: interpretive methods, meta-ethnogrpahy, metasynthesis.

Introduction

The concept of quality of life (QoL) is important for children and young people (CYP) with brain tumors (BTs) in a number of ways. It has figured into outcomes research, into the development of interventions and in clinical trials all of which contribute to the protocols and standards of care used for delivering treatment to these children. Perhaps most important is that it may be used in the decision-making process between CYP, parents and health care professionals (HCPs). Expected QoL is often a key factor in determining whether patients with a life-threatening illness (LTI) or life-limiting condition (LLC) will be given a specific treatment or not (Crane, Haase, & Hickman, 2018; Pellegrino, 2000). This is particularly important in CYP with BTs for whom there are a number of diagnoses, which have a poor prognosis and so have to choose from a number of experimental therapies. Understanding the CYP's perspective of QoL, as well as that of their parent and HCP, is critical to the success of decision making as a joint venture, especially because parents and HCPs may perceive QoL differently (Janse, Uiterwaal, Gemke, Kimpen, & Sinnema, 2005). Recent movements at the turn of the century to include the child's perspective in their care and treatment (House of Commons Health Committee, 1997), makes understanding the views of CYP even more pertinent.

Understanding stakeholders' conceptualizations of QoL is important not just for this application to clinical

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decision making but to all uses of the concept. These concepts are key to building an adequate theoretical or conceptual framework, which guides the development of QoL instruments. As Haase, Heiney, Ruccione, and Stutzer (1999) stress "without development of theory that is grounded in the experiences of patients, we are likely to continue the 'shopping list' approach to HRQL" (p. 130). A search for the conceptual frameworks involved in pediatric QoL measures revealed that most measures have no conceptual underpinning; studies recommend that pediatric QoL research invest time into theory development and evaluation (Davis et al., 2006). Lawford and Eiser (2001) suggest a theoretical model for QoL would clarify the domains and concepts to be assessed in QoL measures (avoiding need for wide range of current measures involving different domains) and understanding how they are related to one another rather than constituting an unrelated list.

The Review

Aim+

To understand CYP, family members and clinicians' understanding of QoL as they use it in decision making for CYP with BTs through a meta-ethnographic review of research published internationally between 2007 and 2016.

Design

Search Methods

Original peer-reviewed articles presenting research exploring stakeholders' concept of QoL in CYP with BTs are included in this review. The search is limited to papers published since 2007 (January 2007—February 2016) reflecting the survival curve for CYP with BTs, which has plateaued since the 2002-to-2007 survival rate data for central nervous system (CNS) tumors was published (Gatta et al., 2014; Munro, 2014).

A comprehensive search of the literature was undertaken using the following databases: Medline, PsycINFO, Embase, CINAHL, and Web of Science. Three main search components were used to identify all relevant and available literature: (a) CYP, parents, and HCPs; (b) QoL; and (c) BTs with a fourth search string to ensure the focus was on the CYP's QoL (not that of the parent or HCP); (d) CYP. A combination of indexed and free text terms were used to reflect these four components. The MEDLINE search strategy is outlined in the supplemental information.

We included English language studies focusing on CYP (aged 0–24 years) with any type of BT, their parents/guardians, and HCPs in charge of their care.

Studies were excluded which:

1. Did not involve CYP (below 25 years old) or their family or an HCP in charge of their care. Any article with a mixed population, for example involving CYP and adults with BTs, will be excluded unless it is clear that CYPs below 25 years of age were the majority of the sample.
2. Focused on the QoL of the parent or HCP and not of the CYP.
3. Did not include BTs in the sample (if it is a mixed sample with other types of tumor this will still be included if data on CYP with BTs are reported separately).
4. Gave no justification for or did not explore the included dimensions or concept of QoL used in or underlying the article. To be included, studies must state/explore the conceptual underpinnings of their QoL. We excluded studies, which simply fitted participants' views into the domains and items of established QoL measures or which looked to operationalize QoL straight from their data without providing participants' concept of QoL. Applying this criterion required going beyond the language of the articles to understanding at a conceptual level and marks the first application of the translational approach used in this review (see the "Synthesis Methodology" section).

Three reviewers (E.B., E.H., G.B.) screened one third of the abstracts identified and assessed them against the identified exclusion criteria. A second reviewer (J.H.) screened 30% of the abstracts overall to test for inter-rater reliability, calculated at 80% agreement. Papers identified from the initial database searches were screened for duplicates, which were removed. Citations were then screened for relevance and those that did not meet our inclusion criteria were removed. One reviewer (E.B.) screened all full texts and three reviewers (E.H., G.B., J.H.) screened 10% of the full texts each. Any disagreements over studies to be included or excluded were discussed by the reviewers until a consensus was reached.

Search Results

Of the 2,346 papers returned by the online search, six remained after screening (Figure 1, see supplemental information). No studies reporting on clinicians' concepts of QoL were found. One of the six studies included parents who acted both as proxies for their deceased children and expressed their own views.

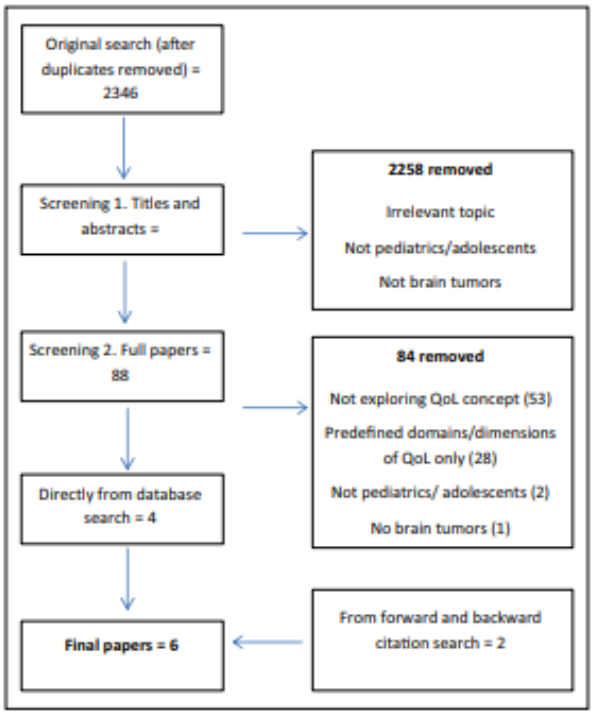

Figure 1. Flow diagram of search and inclusion process.

Quality Appraisal

Guidance for the conduct of systematic reviews recommends the use of quality assessment tools. Included articles were assessed for study quality using two tools. Qualitative research was appraised using recommendations from the Critical Appraisal Skills Programme (CASP) Qualitative Research Checklist (CASP, 2013). Mixed-methods research was appraised using recommendations from Guyatt et al.'s (1993) "Users' Guides to the Medical Literature II." Study quality was assessed for all six studies by two researchers (E.B. and J.H.), and any disagreements were resolved by consensus. Ratings are presented in Table 1. The quality appraisal did not yield any information that has led to the elimination of any of the studies, or to interpret their findings any differently.

Perspectives Represented in the Review

Four studies interviewed YP survivors (Chou & Hunter, 2009; Drew, 2007; Gunn et al., 2016; Hobbie et al., 2016). In one study, both parents and children were interviewed (Darcy, Björk, Enskär, & Knutsson, 2014). Their aim, however, was to capture "children's experiences as reported by themselves and their parents" (Darcy et al., 2014, p. 610), hence it may be taken as providing the views of children with parents as proxies. The children were below 6 years old and undergoing treatment. Zelcer, Cataudella, Cairney, and Banister (2010) interview bereaved parents both as proxies for the deceased CYP and also as reporting on their own experiences. The findings taken from Zelcer et al. (2010) are limited to ones, which relate to the CYPs experiences rather than to parent's own issues, for example, competing priorities and choosing place of death.

Synthesis Methodology

The synthesis employs the method of meta-ethnography, an interpretive form of analysis developed by Noblit and Hare (1988). The fundamental activity of this approach is translation, attempting to "transform interpretations offered by individual studies in such a way that they can be expressed in each other's terms, thereby enabling a direct comparison of seemingly distinctive pieces of evidence" (Pope, Mays, & Popay, 2007, p. 75–76). We have chosen meta-ethnography as our approach to synthesis for a number of reasons: Although some of the studies in the sample are mixed method, the data which concern us are all qualitative; the sample is small and therefore translating the concepts from each study into the others is a manageable task. In addition, the literature reviewed is relatively coherent. Dixon-Woods et al. (2006) identify three basic strategies in meta-ethnography (Box 1). In this review, we employ two of them, reciprocal translational analysis (RTA) and line of argument.

Box 1. Three Major Strategies of Meta-Ethnography (Dixon-Woods et al., 2006).

1. Reciprocal translational analysis (RTA): The key metaphors, themes, or concepts in each study report are identified. An attempt is then made to translate the concepts into each other. Judgments about the ability of the concept of one study to capture concepts from others are based on attributes of the themes themselves, and the concept that is "most adequate" is chosen.
2. Refutational synthesis: Contradictions between the study reports are characterized, and an attempt is made to explain them.
3. Lines-of-argument synthesis (LOA): It involves building a general interpretation grounded in the findings of the separate studies. The themes or categories that are most powerful in representing the entire data set are identified by constant comparisons between individual accounts.

For all studies, we had to identify the key related concepts from the studies by reading closely their reflections on what life is like for the CYP. Identifying related themes and concepts in different studies is an integral part of synthesizing the literature: "studies focusing on similar

Table 1. Study and Patient Characteristics.

Reference	Sample	Perspective	Country	Data Collection Methods (Qualitative Only)	Data Analysis Methods	Quality Appraisal[a]
1. Chou and Hunter (2009)	Young adult survivors (18–21 years old) *n* = 98 (50% with BT 50% leukemia)	YP	Taiwan	Semistructured qualitative interviews (including one focus group) with subsample of participants to further explore QoL and their answers from the questionnaires.	Thematic analysis.	Medium
2. Darcy, Björk, Enskär, and Knutsson (2014)	Children with cancer 1–5 years old. (*n* = 13, four of whom had a "brain or other solid tumor") and their parents.	CYP and parents	Sweden	Semistructured interviews.	Content analysis with an inductive approach.	High
3. Drew (2007)	Young adult survivors (17–27 years old) *n* = 57 (11 of 57 had BT).	CYP	Australia	Qualitative open-ended questionnaires (including "QoL scales") and "in-depth" qualitative interviews with most of sample who answered questionnaire.	A combination of grounded-theory techniques and narrative analysis.	High
4. Gunn et al. (2016)	CYP BT survivors (14–35 year olds, median age = 24 years) *n* = 21	CYP	Finland	Semistructured interview using the phenomenological method.	Thematic analysis.	High
5. Hobbie et al. (2016)	CYP survivors (15–36 years old, *M* = 23) *n* = 41 (all BTs)	CYP	The United States	Cross-sectional, semistructured interview.	Directed content analysis.	High
6. Zelcer, Cataudella, Cairney, and Banister (2010)	Parents (*n* = 25) of CYP (*n* = 17) who had died from BT. All CYP were aged between 1 year and 19 years at the date of death.	Parents	Canada	Semistructured focus group.	Thematic analysis cite Braun and Clarke (2006)	Medium

Note. BT = brain tumor; YP = young people; QoL = quality of life; CYP = children and young people.
[a]We attributed a score to each study corresponding to the checklists we used and then gave each a rating of high (7–10), medium (4–6), or low (0–3). This was not intended to be used to exclude papers but to give the reader some indication of quality of the included studies.

topics may have conceptual overlaps, even if these are not apparent from the way the results are reported" (Popay et al., 2006, p. 18). Our goal in synthesis was to find these overlaps.

Some of the studies reviewed are mixed-methods studies; however, we are using only the qualitative data presented in them; data which are text-based and derived from semistructured interviews or focus groups. We further restrict our focus to the portions of the articles, which are relevant to stakeholder's concepts of QoL for CYP with BTs. This extraction of relevant findings from the studies was itself an interpretative task. This review is thus carried out not simply using an interpretive method of synthesis but from an interpretive perspective throughout. Dixon-Woods et al. (2006) highlight the difficulties of using conventional search strategies for an interpretive synthesis. We extended the use of interpretative methodology to the searching and screening process to alleviate this problem (exclusion criterion 4, above). Even at the stage of screening, we needed to move beyond the literal

Table 2. Overarching Key Concepts Related to QoL for Each of the Included Studies.

Studies	Chou and Hunter (2009)	Darcy, Björk, Enskär, and Knutsson (2014)	Drew (2007)	Gunn et al. (2016)	Hobbie et al. (2016)	Zelcer, Cataudella, Cairney, and Banister (2010)
Key overarching concepts	Personal control (study's overarching concept)	A strive [sic] for normal life (study's overarching concept)	Striving for normal life (we have taken this as their main concept of QoL as they do not present an overarching concept although they frame the experiences as having an emphasis on returning to normal life once participants were disease-free.)	A new normal (two of the study's themes combined [changed health and need for normal] to reach an overarching concept)	Struggle for normalcy (taken study's key theme as overarching concept of QoL)	Striving to maintain normality (one of the study's themes taken as overarching concept)

Note. QoL = quality of life.

text to determine whether we could find a concept of QoL independent of a measure.

Two reviewers were involved in the analysis and translations of the data (E.B. and R.L.). Both reviewers extracted the data and then compared the translations and any disagreements were discussed and a consensus reached.

Results

Synthesis

Key concepts were extracted from each paper and put into tables to understand the concepts of QoL presented by each paper and to allow the authors to interpret and translate the concepts into one another using meta-ethnography (Tables 2 and 3.).

RTA

The first task of analysis was to lay out the primary or overarching concept in each paper that was relevant to our research question (Table 2). To be explicit about how the concepts compared with one another, we created a spreadsheet into which we placed the concepts from each paper.

In five studies we found an overarching concept of "normalcy" or variants (Darcy et al., 2014; Drew, 2007; Gunn et al., 2016; Hobbie et al., 2016; Zelcer et al., 2010). Chou and Hunter (2009) present "personal control" as their main concept. Additional key concepts were identified from each paper to compare and translate the studies into one another. The additional concepts taken from the included studies were independence, social functioning, future, success, physical factors, change, and resilience.

We then created Table 3 which we used to translate the concepts into one another, showing how in each paper the relevant concepts overlapped or differed and explained these (Table 3). From this translation process, we arrived at the conclusion that the difference in overarching concepts of normalcy and control is a difference of point of view, of being process oriented or outcome oriented, control being the process and normalcy being the outcome. So, this dyad of "control-normalcy," process–outcome is the overarching complementary conceptual dyad, which captures a QoL concept underlying all the studies reviewed. Four additional concepts emerged, which provide content to the idea of a new normalcy: personal relationships, independence, success, and a future. These are not simply measurable properties; rather, they identify domains within which survivors must achieve adaptations to realize normalcy and QoL.

In pursuing stakeholders' concepts of QoL normalcy—a new normal—emerges as the key concept with control alongside as a process to reach and maintain normalcy. In the studies in which normalcy features as the overriding concept there are a number of references to striving for normalcy, clearly joining normalcy to process and to activity (Darcy et al., 2014; Drew, 2007; Gunn et al., 2016; Hobbie et al., 2016; Zelcer et al., 2010). Control is not an abstract concept of keeping one's life in order. In the study in which it features it is control with a specific purpose: to bring about those things in one's life which constitute a normal life under circumstances changed by cancer—a new normal (Chou & Hunter, 2009).

We propose then that QoL is something which BT survivors experience in the process of, during the activity of realizing a newly normal life. It is something they experience as a result of the actions which they take.

Table 3. Translations of Key Concepts in Included Studies.

Normalcy

Reciprocal translation:

Five studies highlight the importance of striving for normalcy to these CYP and their parents (Darcy, Björk, Enskär, & Knutsson, 2014; Drew, 2007; Gunn et al., 2016; Hobbie et al., 2016; Zelcer, Cataudella, Cairney, & Banister, 2010). It appears strongly throughout to be a key factor in their QoL. It is expressed in very similar terms: striving for, struggling for, being, a need for "normal," "normality," and "normalcy." A female survivor in Drew (2007) upon reading the experience of another survivor claimed it was the:

> first time in 8 years that she felt she might actually be normal.

Chou and Hunter's (2009) are the only study not to use the terms "normalcy" or "normal life" in their study. Their overarching concept is a process: controlling. However, one can relate Chou and Hunter (2009) to the other studies by seeing their subthemes of "self-sufficiency," "having relationships," and "success" as elements of a normal life. In striving for this combination of elements in their lives they are striving for what the other studies indicate under the single term normalcy.

Chou and Hunter (2009) also relate to other studies in that the object of the control which their participant's value is essentially what the other studies describe as normalcy. We understand in part what the control which they are talking about by the result, by the state which control aims to bring about. The goal of control in Chou and Hunter (2009) is to achieve a state, which substantially overlaps with what is meant by normalcy in the other studies. Conversely, people strive for and achieve an "everyday life" through certain techniques or processes such as Darcy et al. (2014) have as one of their subthemes, "a striving for control." So control and striving for normalcy are arguably accounts of the same reality from different points of view, one focusing more on process (control) and the other on the outcome of the process (normalcy).

CONCLUSION:

Normalcy and the struggle or process to achieve normalcy is present in all six studies and is a key factor in QoL for ill CYP, survivors, and bereaved parents.

Control

Reciprocal translation:

Chou and Hunter (2009) find a theme of control and Darcy et al. (2014) find a theme of striving for control in their concepts of QoL:

> I want to know, I want to do it!

The other four papers (Drew, 2007; Gunn et al., 2016; Hobbie et al., 2016; Zelcer et al., 2010) show a desire for control or affirm the importance of control. Drew (2007) highlights the YP's desire for information that would "promote their endeavors at self-understanding and support their efforts at making the most of their lives after cancer" and their need to manage uncertainties, both of which are ways of asserting control in life. Gunn et al. (2016) express the YP's need for a home and coping on their own as important which gives control back to the YP. Also the expression about uncertainty, similarly to the CYP in Drew (2007), indicates a lack of control and a concern about control. Hobbie et al. (2016) and Zelcer et al. (2010) also touch upon control more indirectly by talking about the dependence on parents and loss of functions and thus indicating the importance of this lack of control.

CONCLUSION:

Control, whether striving for it or expressing a lack of control, seems to feature in most of the CYP's lives and therefore is a component in their QoL concept.

Independence

Reciprocal translation:

There is clear agreement in four of the studies (Chou & Hunter, 2009; Darcy et al., 2014; Gunn et al., 2016; Hobbie et al., 2016), which all clearly present themes of independence as important in the CYP's QoL. This is expressed as self-sufficiency, describing and relishing in self-care activities, and independent living as a goal in both Gunn et al. (2016) and Hobbie et al. (2016). Hobbie et al. (2016) comment how "almost every survivor acknowledged the support of their primary caregiver" and cites a survivors experience of striving for independence:

> I don't always need the help. If you're willing to help me, great, but I don't always need it . . .

The other two studies, Drew (2007) and Zelcer et al. (2010), do not refer literally to independence or dependence in their accounts of CYP's QoL but issues of dependence are implied in or entailed by what they find. Zelcer et al. (2010) give the reader a feel for the parents and CYP's lives through the narratives, themes, and quotes and increasingly toward the end of life authors reported that the parents would have done everything for their CYP and so they were very dependent on their parents. Drew (2007) does not mention independence as a factor but highlights the importance of activities, which exemplify independence as in "Alice, who was just about to travel internationally to continue tertiary study . . ." and so independence is indirectly referred to.

CONCLUSION:

Independence is a key feature in the QoL concept, or striving for independence. And those whose QoL is poor, often depend substantially on their carers.

(continued)

Table 3. (continued)

Social functioning

Reciprocal translation:

There is agreement throughout all of the six studies over the involvement of a form of social functioning. Chou and Hunter (2009), Drew (2007), and Gunn et al. (2016) express this as relationships and Zelcer et al. (2010) as "friendships" and "support of peers," which they report was of significance in the adolescent population. Hobbie et al. (2016) refer to it directly as social functioning, and difficulties in social functioning were demonstrated by one survivor:

> The hardest thing in my life is getting along with my friends. . . people I work with. . . I want to have friends, but I just can't seem to find them.

Darcy et al. (2014) stress the CYP's simple need as a "longing for others." Zelcer et al. (2010) also highlight the loss of the CYP's ability to communicate which in turn affects their social functioning.

The fact that all six studies directly refer to forms of social functioning and some even stress it as the most affected or important of the CYP's lives further supports using an interactionist perspective to analyze the studies as QoL seems heavily weighted toward relational context.

CONCLUSION:

Social functioning or relationships feature in all six studies directly and a key component of QoL.

Future

Reciprocal translation:

Two studies need no translation or interpretation and are in agreement over inclusion of the term "future" when describing what is important to the CYP. Chou and Hunter (2009) talking about a future orientation and having hope. Drew (2007) expresses the YP's tension over past and future experience, the survivors felt a need to have knowledge and details about their illness history to best conceptualize and plan for the future as demonstrated by one male survivor:

> They're very bad with that information. [The fertility clinic] sent me a letter two years ago saying no storages . . .(are). . . allowed to be kept for more than ten years. . . I contacted them and said "I know it's a few years coming, but you're not going to throw those out are you." . . .

There are also indirect ways in which the studies express the future through the wishes and aims of the CYP in Hobbie et al. (2016) to one day date or live independently. This is an indirect way of articulating the need to aim for something in the future. Zelcer et al. (2010) similar to Chou and Hunter (2009), talk about maintaining hope which is about anticipating or conveying the future.

Gunn et al. (2016), however, has a theme "living one day at a time," which indicates a different approach to a future. Darcy et al. (2014) also indirectly speak more to this lack of future by emphasizing the focus on everyday life with no future scenarios mentioned in the narratives.

CONCLUSION:

All six studies recognize that BT survivors must deal with the issue of a future in achieving a new normal and adapt it to their circumstances. Some studies feature the future explicitly as a component of the QoL concept, including things such as hope. Others focus more on living in the present and future. In both sorts of cases the future must be dealt with and redefined in a way compatible with changed circumstances.

Success

Reciprocal translation:

Only Chou and Hunter (2009) use the word success in their concept of QoL. In four of the other five "success" is alluded to indirectly by talking about impact of the disease on education (Hobbie et al., 2016; Zelcer et al., 2010); education and employment (Drew, 2007) and vocation/education as a female survivor explains:

> comprehensive school went just alright, because I got so much support. . . (Gunn et al., 2016)

Darcy et al. (2014) do not touch upon success directly; this could be due to the child's age as most were infants or very young children who were not old enough for school. Preschool was touched upon but only related to the social issues of preschool, not related to the success of attending. Success was inferred very indirectly through accomplishment of gaining knowledge and understanding, being participatory in their care to the degree that they wanted to, and feeling safe and secure in parental presence. Similarly to normalcy, success is the end result of the control and so success and control are both partial accounts of the same phenomenon.

CONCLUSION:

Some form of success is featured in most of the studies (five/six) with mentions of educational or vocational success being present, but seemingly not key, in the concepts of QoL (Chou & Hunter, 2009; Drew, 2007; Gunn et al., 2016; Hobbie et al., 2016; Zelcer et al., 2010).

(continued)

Table 3. (continued)

Physical factors

All six studies mention some aspect of physical factors.

Drew (2007) and Hobbie et al. (2016) both described how physical aspects of the illness affected their appearance, Drew (2007) highlighting how this in turn affected their self-consciousness:

> The cancer has left me in a wheelchair for eighty percent of the time. So sometimes I feel really angry, and other times I just accept it as part of my life.

Hobbie et al. (2016) commented on the number of respondents dealing directly with late effects of disease and treatment. The parents in Zelcer et al. (2010) emphasize how the neurological deterioration affected how they could partake in activities they enjoyed most.

Darcy et al. (2014) have a theme "living with a changed body," but it seems children learned to adapt to the illness and its effects quickly. Similar to Zelcer et al. (2010), the way the illness affected the CYP was through impact on activities rather than illness itself.

However, Chou and Hunter (2009) stress that it is the psychosocial not the physical aspects that affect on a CYP's QoL. Gunn et al. (2016) too, highlight that despite many physical symptoms disclosed by the CYP they reported themselves as healthy. This is in line with the response shift theory, which states that changes in self-response measures occur over time as a result of recalibration, reconceptualization, and reprioritization of internal standards.

CONCLUSION

In half the studies (3/6) physical factors seem to impact directly on the CYP's QoL and therefore feature in a QoL concept (Drew, 2007; Hobbie et al., 2016; Zelcer et al., 2010). It mostly seems to affects the way the CYP can take part in usual activities (and therefore interfere with their normal everyday life). However, half of the studies state explicitly that it does not feature in the QoL concept and emphasize the ability of the CYP to adapt to the illness and find a new normal (Chou & Hunter, 2009; Darcy et al., 2014; Gunn et al., 2016).

Change

Reciprocal translation:

Four of the studies include an aspect of change in their discussion of QoL. Darcy et al. (2014) have themes of a changed body and living in a changed home. Drew (2007) focuses on changes to the YP's identity and self-concept. Gunn et al. (2016) have themes of positive change and a changed understanding of the term health. As one male survivor put it:

> I kind of understood that you can also gain from negative things, or they are part of life and you should enjoy them in a way and learn, and not just get rid of them

Hobbie et al. (2016) emphasize the struggle for normalcy among all the physical, social, cognitive, and emotional changes occurring in the CYP.

Chou and Hunter (2009) and Zelcer et al. (2010), however, have nothing emerging directly on change from their studies. But indirectly by talking about the loss of functions and ability to communicate this indicates change implicitly. And similarly Chou and Hunter (2009) talk about striving for control and striving indicates change is needed and aimed for. With Zelcer et al. (2010), the lack of aspects of change may be a reflection of the retrospective methods of data collection. Asking parents many years after the experience rather than when they are in the experience in real time may not be able to recall changes that were occurring at the time.

CONCLUSION:

Change features directly in most of the studies (4/6)(Darcy et al., 2014; Drew, 2007; Gunn et al., 2016; Hobbie et al., 2016) with references to changed bodies, self-concepts and this idea of generally adapting to new circumstances occurring in others (Chou & Hunter, 2009; Zelcer et al., 2010).

Resilience

Reciprocal translation:

Only two studies directly mention resilience. Zelcer et al. (2010) have resilience as a subtheme of sources of spiritual strength as one mother explained:

> We were surrounded with love. He knew that; he was so good, he directed his own care and directed us and he had good quality of life.

They stress that parents talked about how strong and resilient the CYP was throughout their illness. It does not feature in Chou and Hunter (2009)'s concept of QoL, but they present QoL as part of a bigger concept/model called the adolescent resilience model and in this QoL is seen as an outcome from the resilience that CYP have.

The other four studies encompass some form of resilience indirectly whether mentioning the CYP's ability to bounce back (Darcy et al., 2014) becoming "stronger" (Drew, 2007), an increased positive attitude (Gunn et al., 2016) or wanting to give back as they had become disease free themselves (Hobbie et al., 2016). Drew (2007) and Gunn et al. (2016) also highlight the CYP's gratitude for just being alive.

The use of the word resilience in Zelcer et al. (2010) may also be a reflection of the method and speaking to the parents rather than the CYP themselves as it is a reflection about strength of character that is easier to express in hindsight looking back over a period and also about another person rather than oneself.

CONCLUSION:

Having resilience is mentioned either directly or indirectly in all six studies. This idea of the YP gaining something positive from the diagnosis features in most of the studies and seems to be a part of the QoL concept.

Note. CYP = children and young people; YP = young people; QoL = quality of life.

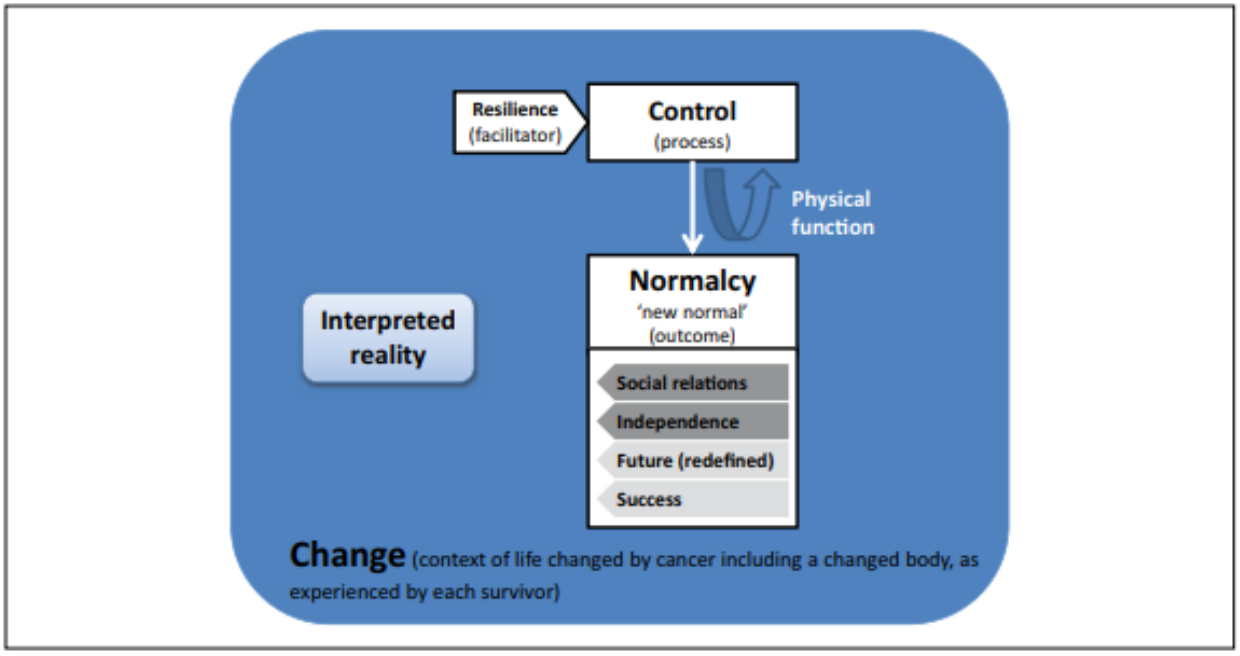

Figure 2. Model of quality of life.

What this indicates that (new) normalcy is not a "static" property as an object's being of a certain size, shape, or color. Experiencing a measure of (new) normalcy is an achievement which needs to be constantly repeated. Because normalcy is something which applies to one's life because of activity, we present the concepts of (new) normalcy and control as a dyad.

Line of Argument: A Conceptual Model of QoL as Striving for a New Normal

Three further concepts emerge from our studies: resilience, physical function, and change. These are not components or dimensions of QoL, of what that means to CYP but they nevertheless have fundamental relations to QoL and how that achievement comes about. These three play different roles than do the first six (normalcy, control, independence, social functioning, future, and success) and can be fitted with them to form the beginning of a model in which our concept of QoL is situated.

Change is a fundamental aspect of survivors' experience and is the context within which they seek and achieve a new normalcy. They experience a changed body and changed relationships. Changes in physical functioning—basic physical abilities and cognitive function—are key elements in this context of change. Again, these are not items which can be straightforwardly measured and compared between individual survivors. These affect survivors' QoL as they are experienced and interpreted by the individual. Thus, what an external observer might call the same change or degree of change can be interpreted differently by different individuals and thus have a different impact on QoL.

Physical functioning (including neurocognitive impairments) is thus not a part of survivors' QoL concept, but is something to which BT survivors adapt. It can be seen as something which pushes back against, offers resistance to the control element. These elements are all set against the arena of "change." All studies reported that CYP with BTs go through some form of change, internal (e.g., physically, emotionally) and external (e.g., in the home, educationally), and this changed life is the context within which CYP strive to achieve a new normalcy. So set against this arena of change the central concept of QoL can be seen to be this idea of "new normal," striving for a new type of normalcy (Figure 2).

Resilience is something all CYP have in differing degrees and can be seen as influencing the amount of control a CYP has to achieve normalcy. It reflects survivor's ability to adapt to change and interpret changes in function in a way which allows them to be integrated into a new normal.

Discussion

Key Findings

This review explores the concept of QoL in CYP with BTs from the perspective of two of the stakeholders in decision making about care and treatment. Meta-ethnography is used to synthesize the six studies identified as presenting

findings relevant to a concept of QoL in CYP with BTs from a stakeholder perspective. Striving for a new normal is the core concept to come from the synthesis. A related concept, control can be seen as an essentially linked process, as behavior strategically aimed at achieving normalcy. Resilience facilitates control. Elements of social relations/functioning, success, independence and a redefined future emerge as key aspects of the concept of a new normalcy. A changed functioning, which typically includes deficits in neurocognitive functioning, is something which pushes back against or offers resistance to the element of control. These elements are all set within the arena of "change," a life changed by cancer.

Our Findings and Previous Literature

Previous studies of CYP and families experience with cancer have found striving for normalcy and the establishment of a new normal as a key component of living with and after cancer and chronic illness. Clarke-Steffen (1997) find that life with and after cancer was different but assumed a quality of normality. Parents and CYP had a new "world view." Stewart (2003) in a study of younger (preadolescent) children undergoing treatment for cancer found that they viewed their lives as routine and ordinary. In reviewing literature on the concept of normalization, Deatrick, Knafl, and Murphy-Moore (1999) report that in normalization, change, and impairment are acknowledged. Normalization, they say, involves both cognitive (definitional) and behavioral (strategies) process and that these two aspects are "inextricably linked." Taylor, Gibson, and Franck (2008) find that "Young people with a chronic illness strove to overcome social, psychological and physical difficulties to have a normal life" (p. 1828). Earle, Clarke, Eiser, and Sheppard (2007) find that the mothers of CYP with acute lymphoblastic leukemia in their study were all striving to achieve a normal life and had to adjust to a new normal since the diagnosis. Van Schoors et al. (2018) also found families with a CYP with leukemia (or non-Hodgkin lymphoma) were striving for normalcy. In a study of children with developmental delay and cognitive impairment but from illnesses other than cancer, Rehm and Bradley (2005) find that some parents rejected the description of their lives as normal in recognition of the differences in their lives from those of normal families to which they compared themselves and in which there was no illness. Other families characterized their lives as a "crazy normal" or said they were "normal but . . . ."

What this review shows is that achieving this combination of redefinition and normalizing practices brings a sense of quality into the lives of YP and, we suspect, their families as well. QoL is something which results from a combination of cognitive, or meaning making, and behavioral strategies but is not identifiable with these individually. As Deatrick et al. (1999) write "No one attribute can be considered without viewing its context and the attributes as a set. While attempting to clarify the conceptual foundation of normalization, defining attributes are necessarily reductionistic" (p. 213).

In a study published prior to the dates of our search, Hinds et al. (2004) propose a definition of QoL for CYP with cancer based upon interviews with CYP. They define QoL as "an overall sense of well-being based on being able to participate in usual activities; to interact with others and feel cared about; to cope with uncomfortable physical, emotional, and cognitive reactions; and to find meaning in the illness experience" (p. 767). Comparison with our proposed concept (survivors' concept) is useful and brings to light some basic issues encountered in efforts to define or conceptualize QoL for CYP with cancer and with BTs in particular.

Both our concept and Hinds' are holistic. Hinds et al. (2004) builds on an overall perception of the CYP's life; our concept relies upon the YPs overall characterization of their life as normal in a form reassessed post illness. An interesting point of difference is that symptoms and physical function do form a part of Hinds' concept of QoL whereas from our concept they are part of the model but not of the concept per se. Significantly, in Hinds' concept the impact of symptoms and function is mediated through the YPs ability to cope with these changes in the YPs life. Thus, on both views the impact of physical changes is not direct, and do not lead inevitably to a decreased QoL. Furthermore, by featuring adaptation, or in our case resilience as well, both concepts allow for positive gains after or during an experience with cancer.

In making this comparison, it is important to note that Hinds et al. (2004) observe that a limitation of their definition is that it is based solely upon views of CYP currently receiving treatment and included no long-term survivors. Our study suffers from a similar type of limitation but in precisely the opposite direction: it is weighted toward the views of survivors.

From the Concept of QoL to Measurement

Good measures of QoL, in addition to having sound psychometric properties, need to reflect issues which are important to YP and families (Eiser, 2004; Hinds, 2010). Despite this agreement in the literature that a need for normality is what families want and how they understand well-being, measures of QoL do not feature normalcy and achievements. Instead they measure deficits in several domains. As Haase et al. (1999) stress, this is a limited way of assessing QoL and without assessing the meaning for the individual CYP. No measure focuses on how far they can live their normal everyday life. The European Quality

of Life-5 Dimensions (EQ5D; The EuroQol Group, 1990) approaches this with a domain of how able a patient is able to undertake their "usual activities" be it school, study, or work and the Child Health Utility instrument (CHU9D; Stevens, 2009) with the domain "able to join in activities" but these are one of many domains in the measures.

Our review stresses the primacy of social relationships in achieving normalcy and by inference a good QoL. Hobbie et al. (2016) stress how the importance of families went beyond the resources, structure, and support of their emotional, physical (etc.) functioning and "provided the recognition that they were important beings and their existence mattered to someone" (p. 140, but also featuring in the abstract). They describe the family as a "looking glass," which enabled survivors to define themselves and enjoy a positive sense of self. Instruments which are sensitive to the concerns of YP and families need to address these aspects of CYP's concept of QoL.

Different Agendas

It is important to keep in mind that different approaches to QoL, qualitative approaches exploring individual's appraisal of their QoL versus quantitative approaches using instruments, typically have different purposes. Qualitative studies are important for HCP in their clinical interactions with BT patients and families. Many quantitative studies of QoL are conducted to document a need for continued support for BT survivors. To justify support services, these services need to have measurable impact. Haase et al. (1999) talk of the "pressing need to measure HRQoL as an outcome for children and adolescents" while at the same time they recognize that this "seems to be leading to an acceptance of function-based models without a critical evaluation of the underlying assumptions for both function and meaning as the basis for HRQoL assessment" (p. 125).

This review was undertaken, by contrast, to understand how participants in clinical decision making might understand and use the concept of QoL in those discussions. We are not focused on issues of defining needs for a population and measuring change in QoL over time.

Strengths and Limitations

Although we feel confident about our application of the principles of translational, meta-ethnographic, synthesis to the studies reviewed, we are nevertheless aware of a number of ways in which our results, conclusions, and hypotheses should be qualified. As with any review our results are limited by what was returned by the search. Our search yielded nothing on clinicians' understanding of or use of the idea of QoL and virtually nothing about parents'.

Each study is from a different country with varying health care systems; one was from a non-Western country. Nevertheless, there was no opportunity to explore the possible impact of social factors at a macro level on understandings of QoL. Such differences may exist within Europe. Radiotherapy is devastating to a young, developing brain. Guidelines about the minimum age at which it should be used vary across different countries in Europe. In the United Kingdom, radiotherapy is avoided in cases of children below 3 years of age. In France, the recommendation is that be avoided until 5 years of age. There is some speculation that this reflects societal attitudes toward disability.

Our approach to these studies has been interactionist, a more microsociological approach, and focused on BT survivors interacting with family and friends to create a new normal life. We recognize that social actors do not create social life ex nihilo. Our analysis can be complemented by analysis from other perspectives, by showing how larger scale social factors influence and support this interactional reality.

Samples in the studies reviewed were purposive or convenience samples; rates of participation of eligible subjects, when they could be calculated (four studies) ranged from 48% to 32%. We cannot know the reasons why a substantial number of eligible participants either refused to participate or simply neglected to do so and whether these decisions or events had an impact on our "data." It is possible that those most severely affected by treatment for BT more often chose not to participate than other survivors.

We also lack demographic information on participants. Economic resources of the families might affect on the ability of survivors to adapt in these challenging circumstances.

Reporting on the treatment stage of the CYP is a critical component of analyzing a CYP's experience yet it is regularly ignored in research (Taylor et al., 2008). QoL is dynamic and likely dependent on where in the illness trajectory the CYP is. Hinds (2010) stresses that QoL can vary as severity of treatment changes and continue to do after treatment ends. Our small cohort of studies is composed largely of survivors (213 of the 243 total subjects) some years after diagnosis/treatment. Only Darcy et al. (2014) had CYP participants undergoing active treatment, although nothing they found there conflicted with the other studies. Exploring the concept of QoL of CYP's who are undergoing active treatment is an area that warrants further investigation.

Conclusion

The clear finding of this review is that BT survivors' understanding of QoL is based upon adaptation to their

changed circumstances. This stands in contrast to a deficit approach to QoL which is typically found in QoL scales and measures. How might the differences between the two approaches be resolved? Should one or the other be regarded as the better understanding of QoL? What are the possible consequences and implications of this difference for CYP with BTs?

Decisions about interventions and treatment protocols may be being made for this population by clinicians and researchers, relying upon deficit-based instruments, whose assessment of the QoL of BT patients and survivors may differ from those of the patients themselves and of their families. Researchers may regard proposed new therapies as resulting in a low HRQoL but patients and families might judge the outcomes to be acceptable. This could slow progress in improving outcomes-survival for BT patients. But, allowing therapies with a higher burden of long-term effects places a new burden on parents and CYP as decision makers. To take such risks, decision makers must clearly envision what survival would be like and what their ability to adapt to and enjoy such a life would actually be. This is something which CYP and their families must do in a context, which is emotionally charged and often beset by uncertainty. As noted above, the understanding of QoL developed here may not reflect the experiences of survivors who have suffered the greatest neurocognitive damage.

One of the uses of deficit-based HRQoL instruments is to document the need for long-term surveillance of BT survivors and the provision of support services. The findings in this review could be taken as presenting advocates for survivors with a dilemma of either renaming their instruments as health status scales or relying more heavily on patient and family subjective assessment of their QoL. However, separating their instruments from the idea of QoL risks making their petitions to policy makers and commissioners for services for BT survivors seem less compelling, about something less important, especially if it is thought that survivors and their families assess their QoL as at least acceptable.

At the same time, it needs to be recognized that while deficit-based instruments may be effective in securing support for BT survivors, they may not be well suited to planning and guiding the services which survivors actually want and need. The adaptive concept of QoL presented here may be better for those tasks. BT survivors find QoL within the milieu which they create with other members of their family. Services need to support this unique unit. Assessments of needs based on external and universal criteria may not be suitable for doing this.

Health-related quality of life (HRQoL) measures have and will continue to have an important role in the lives of BT patients and survivors. Perhaps one outcome of this brief discussion is that it needs to be kept in mind that quantitative measures are instruments, means to ends and not normative judgments.

Clinical Implications

In decision making, support must be provided to clinicians to discuss QoL in understandable terms and whose meaning is shared by CYP and families. Perhaps rather than fixing on the term QoL, clinicians should ask what is important to CYP and their parents and what are their long-term goals.

Clinicians dealing with CYP and families who are undergoing or have completed therapy should be aware of families' struggle to contain the intrusion of the illness and to maintain normalcy as far as possible. They need to accept that normality and independence are subjective notions and mean different things to different people, CYP and their parents. Excessive emphasis on health status and physical functioning can lead to a neglect of patients' and families' understanding of QoL.

Care and service planning should be guided at least as much by what is learned from explorations of the social and subjective dimensions of survivors' QoL as from studies of the deficits revealed by the application of HRQoL instruments.

Directions for Future Research

This study highlights how little is known about the individual stakeholders' concepts and understandings of QoL. This is the case even though the terms are often used in clinical consultations in which decisions about care and treatment of YP are made. It is crucial to investigate how different stakeholders in the clinical consultation understand QoL if there is to be clear and meaningful communication between them.

Clinicians are trained to assess patients objectively typically using standardized quantitative instruments. Hence, one might suspect that they place great emphasis on health status and physical function in thinking about QoL, not just HRQoL. We need to determine whether this is the case and what its consequences might be for interacting with CYP and families, and on decision making.

Parents' understandings of QoL are just as important. Many BTs occur in children below 5 years of age and so have a very limited role in decision making. In such cases, parents become the primary decision makers together with the clinician.

There is a need for an ethnographic longitudinal study that follows families in consultations throughout their illness trajectory to understand what they mean, when QoL is discussed in these consultations. We need to see what is important to CYP and their families and how this changes over time and as the disease changes. Only then will we

begin to understand how each of the three stakeholders conceptualizes QoL and how it functions in clinical decision making. This is as essential for designing effective interventions to improve the lives of CYP with BTs as is looking at it from a deficits perspective.

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Supplemental Material

Supplemental material for this article is available online.

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APPENDIX 4 – HRBT PROJECT ONE PAGE RESEARCH PROJECT OVERVIEW

Understanding decision making for children with high-risk brain tumours: A Prospective, Longitudinal Study of Parents, Children and Clinicians to Provide Guidance for Clinical Consultations

Research aims

- To inform the development of evidence-based guidance which will assist parents, children and clinicians in decision making regarding care treatment and research participation.
- To provide a robust description and analysis of decision making for children with high-risk brain tumours over the entire course of the illness with attention to the views, perspectives and actions of parents, children and clinicians.

Purpose and Design

This project seeks to understand decision making for children with a very poor prognosis. A full and complete understanding of the decision-making process requires attention to all of the parties involved and their interactions with one another over the entire course of the illness.

The research has two interrelated components: (1) A prospective longitudinal study, focused on the interactions amongst, clinicians, parents and children over the entire course of the child's illness and (2) research workshops bringing together all study participants to grapple with the learning from data generated in the longitudinal study and develop from this new knowledge to inform guidance for clinicians, parents and children.

The prospective, longitudinal, ethnographic, participant observation study of children with high-risk brain tumours, their parents and staff involved in their care, will collect data over 20months. Ethnography with its emphasis on immersion in the world of those being studied and analysis of what is said and observed in context provides insights that are otherwise unavailable. The focus will be on the actual interactions/encounters that take place among parents, children and clinicians at all phases of the illness trajectory.

Sample

There are over 400 cases of Central Nervous System (CNS) tumours in children a year in the UK and approximately 20% of these can be regarded as high-risk (median survival < 2 years). The [paediatric hospital] is one the largest paediatric neuro-oncology unit in the UK with 100 new patients a year (~20 high-risk brain tumours). All children treated and cared for at the joint university hospital and tertiary paediatric hospital with high-risk brain tumours (including high-grade glioma, diffuse intrinsic pontine glioma, atypical teratoid rhabdoid tumours and some high-risk CNS- Primitive neuroectodermal tumours (PNET)) will be eligible to join the study at any point in their illness trajectory, from diagnosis, referral to either hospital for further treatment options, participation in research or a second opinion. Based on referral patterns to the study hospitals we anticipate a sample of 16-20 children during 20 months of data collection. The children with high-risk brain tumours are the index cases of our sample.

The children, parents and clinicians involved in their care and treatment will all be research participants in the study as it is our intention to provide a robust description that accounts for each perspective. Clinician participants will include members of neurosurgical, oncology, radiotherapy and palliative care services at the study hospitals involved in their care and treatment. Key staff members will include Neuro-oncology consultants, Neurosurgeons, Radiotherapy consultants, Nurse Practitioners/Specialists in the field of neuro-oncology, Oncology outreach nurse specialists and Palliative Care consultants. In

addition, other clinicians who have significant input into care and treatment decisions for children participating in the study will also be invited to participate. This may include registrars or senior paediatric trainees in any of the disciplines described above and consultants in other specialities such as endocrinology and intensive care.

Core Research Project Team: [removed for identification purposes)

APPENDIX 5- HRBT PROJECT CODEBOOK

[#]	Node	Definitions of node/ Instructions for Coders
N-1	SURGERY	**SURGERY** Discussion about and related to surgical option with regard to: • excision of tumour • debulking • biopsy • cyst aspiration Includes discussions of: • logistics • description of procedure • likely outcomes • risks • benefits • preparation • consent NB: Internalisation of a drain is a shunt and not a surgical procedure. Shunts are dealt with separately in N-9.
N-2	RADIOTHERAPY	**RADIOTHERAPY** Discussions about and related to radiation/radiotherapy option for both disease directed and non-disease directed therapy Includes discussions about (e.g. Milan protocol, COG, ST Jude's, Baby Brain) with regard to: • logistics of Tx • risks • benefits • consent • play therapy • hospital stays • impact on child and family • side effects • efficacy • likely outcomes • general anaesthetic (GA) NB. This also includes the lay language such as 'X-ray treatment'
N-3	CHEMOTHERAPY	**CHEMOTHERAPY** Discussions about and related to chemotherapy options regardless of how that chemotherapy is labelled. Includes: discussions about (e.g. Milan protocol, COG, ST Jude's, Baby Brain) with regard to: • 1st line • 2nd line • Standard • "palliative chemo" • maintenance • high dose chemo with stem cell rescue • oral • IV • adjuvant chemo e.g. methotrexate • adjuvant drugs (non-chemo) • intrathecal chemo-therapy • induction chemotherapy Regarding: • logistics • names of drugs • side effects • efficacy • mode of delivery • risks • benefits NB: Clinical Trials are dealt with separately in N-4 NB: Steroids are dealt with separately in N-6

[#]	Node	Definitions of node/ Instructions for Coders
N-4	CLINICAL TRIALS	**CLINICAL TRIALS** Discussions about and related to clinical trials whether or not they are available. Includes discussions about (e.g. Panobinostat, Sirolimus, Herby) with regards to: • logistics • randomisation • description of agents themselves • risks • benefits • phase I/II/III • compassionate use • therapeutic trial • tissue or bloods needed for trial or research participation
N-5	NAMED PROTOCOLS	**NAMED PROTOCOLS** Discussions about and related to treatment protocols. Includes discussions about the following protocols: • HIT SKK • Baby Brain • High Grade Glioma • St Jude's • Milan • EuroRabdoid (sometimes called EURORHAB) • COG • PVC (also called second line chemotherapy) N.B some of these protocols, as stated, are also clinical trials or types of chemotherapy. Where stated, double code e.g. PVC if referred to as a protocol and as second line chemotherapy, the section will need to be coded as both named protocols and chemotherapy.
N-6	STEROIDS	**STEROIDS** Discussions about and related to steroids Includes discussions of: • logistics • risks • benefits • impact on QOL • dosing of steroids • purpose (e.g. retain swallow, increase appetite, reduce swelling, symptom control, pain management, seizures) • taste • preparation (liquid or tablet NB: Steroids talked about in the study are prednisolone and dexamethasone (also referred to as 'dex')
N-7	OTHER MEDS, MODES OF DELIVERY AND DEVICES	**OTHER MEDS, MODES OF DELIVERY AND FEEDING DEVICES** Discussions about and related to other medicines (excluding chemotherapy and steroids which are covered in nodes N-3 and N-5) modes of delivery and feeding devices including but not limited to: • antibiotics • pain medication • antiemetics • laxatives • hickman • PICC • PEG • NG tube • TPN • Platelets • blood • symptom care plan • anti-seizure meds • fluids • blood products and transfusions • GCSF • bandages • wheelchair • pressure stockings NB: If a parent enquires about any other medication available for their child then code under this node

[#]	Node	Definitions of node/ Instructions for Coders
N-8	COMPLE-MENTARY/ ALTERNATIVE THERAPIES	**COMPLEMENTARY/ALTERNATIVE THERAPIES** Discussions about and related to complementary/alternative therapies including but not limited to: • massage • herbalism • acupuncture • aromatherapy • mindfulness/meditation • cannabis oil/fruit
N-9	SHUNTS AND DRAINS	**SHUNTS AND DRAINS** Discussions about and related to shunts. Includes discussions of: • placement • type of shunt: external (e.g. External ventricular drainage [EVD]) and internal (catheter for brain) • long term consequences • short term consequences • Risks • benefits NB: Internalisation of a drain is a shunt and not a surgical procedure so should be coded under this node and not N-1 Surgery.
N-10	PALLIATIVE CARE	**PALLIATIVE CARE** Discussions about and related only to palliative care explicitly, palliative care services, the palliative care team and its cognates. Includes discussions regarding: • descriptions of what paediatric palliative care is • service provided • attitudes towards palliative care • other services that work with PC team, restricted to: oncology outreach teams, Paediatric oncology outreach nurse specialists (POONs) and symptom care teams NB: Some of these discussions will come up at time of recurrence and take form of "referral". NB: No level of interpretation should be needed to define palliative care. Purely mentions of the above services and the team. NB: If a palliative care team member is involved in a conversation with an oncology consultant about something non-palliative care related this is not to be included. NB: Palliative care does not organise statements of educational needs – this is dealt with by psychology. They do perform school visits but these are to discuss what to do should a child become unwell while in school.
N-11	ACP: ADVANCE CARE PLANNING	**ACP: ADVANCE CARE PLANNING** Discussions about and related to activities and documents that are part of what is sometimes referred to as advanced care planning – emergency care plans, ambulance directives, DNR (may overlap with where it is a specific code linked to a particular context and situation – during surgery) Includes discussions, descriptions of: • DNR/ DNAR • emergency care plans • ambulance directives • place of care NB: When death has been raised directly or indirectly as impacting on place of care • preference in place of care

[#]	Node	Definitions of node/ Instructions for Coders
		• place of death • preference in the place of death • organ donation • autopsy/ post mortem • ventilation
N-12	NOT TREATING THE DISEASE	**NOT TREATING THE DISEASE** Explicit mention/discussion of not treating the disease with surgery, radiation, chemotherapy or clinical trial. This index node is for the overall plan of the sort to treat or not to treat this disease. It is not about a break in treatment. Includes discussions of: • to treat, not to treat • stopping treatment
N-13	SECOND OPINION	**SECOND OPINION** Apply this code to segments of text dealing with obtaining a second opinion on diagnosis or treatment from another doctor/surgeon at any point in the child's illness. The second opinion can be sought via email, face-to-face interaction or phone (but not including an MDT solicitation for an opinion for a case) Including: • from another HCP within the same hospital • from another hospital • from another country
N-14	OTHER SERVICES AND TEAMS WITHIN STUDY HOSPITALS	**OTHER SERVICES AND TEAMS WITHIN STUDY HOSPITALS** Apply this code to segments of text dealing with any other services and teams within study hospitals. Including: • neurology • PICU/NICU • physiotherapy • cardiology • gastroenterology • genetic counselling • psychology • social work **Not** including • radiotherapy • surgery • palliative care • neuro-oncology
N-15	OTHER SERVICES AND TEAMS OUTSIDE STUDY HOSPITALS	**OTHER SERVICES AND TEAMS OUTSIDE STUDY HOSPITALS** Apply this code to segments of text dealing with any other services and teams outside study hospitals. Including: • local hospital teams e.g. Whittington, Newham, Jack's Place • community teams (e.g. Diana, Life-force, Watford) • physio-therapy • hospice (e.g. Chase, Demelza, Richard House, Shooting Star) • local speech and language • Rainbow Trust • GP

[#]	Node	Definitions of node/ Instructions for Coders
N-16	**AIM / PURPOSE / GOAL**	**AIM / PURPOSE / GOAL** Apply this code to segments of text about and related to the intention of the proposed intervention/ option. Includes segments of texts about: • purpose • goal • short term long term • purpose / goal in and of itself or in relation to other purposes or goals (e.g. eligible for another Tx) • buying time • buying time until there is a better alternative available • making another intervention possible • managing symptoms
N-17	**LIKELIHOOD OF ACHIEVING AIM/ PURPOSE /GOAL**	**LIKELIHOOD OF ACHIEVING AIM/ PURPOSE /GOAL** Apply this code to segments of text about and related to the possibility of the proposed intervention succeeding or failing e.g. be of 'benefit'. Possibility can be expressed in any number of ways including: • statistically • likert • statement (e.g. 'in my experience of treating children…') NB: Need to note if the segment of text was non- patient specific (N-19). If we do not see that assume it was pt specific.
N-18	**RISK**	**RISK** Apply this code to segments of text dealing with what could happen, as a result of particular intervention long term and short term – i.e. what would be a bad result of undertaking the intervention. Includes segments about risk of: • death • disability • brain damage • decreased quality of life • possible negative outcomes of undertaking the particular intervention • create new problems (e.g. increase hunger at time when there is no swallow – steroid; extend life when patient no longer conscious -shunt) • side effects • lack of risk
N-19	**NOT PATIENT SPECIFIC**	**NOT PATIENT SPECIFIC** This code should be applied to all segments of texts made that are not about the specific patient (i.e. the child of the parents in the consultation) with regard to the nature, course or Tx of the illness. Includes: • population data • reference to other children • textbook information or history of a disease/treatment • information from the internet • leaflets e.g. from McMillan or Cancer Trust

[#]	Node	Definitions of node/ Instructions for Coders
N-20	**PRACTICAL LOGISTICS**	**PRACTICAL LOGISTICS** This code should be applied to all segments of text related to planning, implementation and co-ordination of any aspect of treatment, tests, social or family life around treatment and scans as well as logistics of living with a brain tumour. It includes but is not limited to: • organisation of the room and who is in it, answering phones, moving chairs etc. • appointment times • location of care and treatment i.e. what will be given at home, what will be given in hospital and what will be given in hospice. • organisation of child care of patient • timing of treatment intervals • delays in treatment, tests or planning, not related to the patient's condition (e.g. no beds, no scanner available) • co-ordination with other teams i.e.hospital teams, CCN teams, local hospital teams, community physiotherapy
N-21	**CONSENT**	**CONSENT** Apply this code to segments of text dealing explicitly with consent to Tx, procedure and research Includes segments of text about: • consent in written or verbal form • explanations of consent procedures • discussions of information sheets
N-22	**THE TUMOUR/ DISEASE**	**THE TUMOUR/DISEASE** Apply this code to segments of text dealing with the tumour as an entity. Includes segments of text mentioning: • location of tumour or metastases • name of tumour • tumour growth/shrinkage/stability • extent of disease • classification of tumour • characterisation of tumour (e.g. aggressive, nasty) • includes saying the C word (e.g. cancer, malignancy) • causes of tumour e.g. pregnancy, genetics, diet • tumour genetics
N-23	**CURE/ CURABILITY**	**CURE/CURABILITY** Apply this code to segments of text where there is explicit mention of the word cure or its cognates (e.g. curative, curative intent, incurable). Includes segments of text dealing with: • potential of the intervention to cure, or in the case of a patient to be cured or in the case of the disease potential of the disease to be cured NB: Does not include 'survive' or 'disease free' [survival] or 'responsive to treatment'.

[#]	Node	Definitions of node/ Instructions for Coders
N-24	**SURVIVAL/ LENGTH OF LIFE**	**SURVIVAL/LENGTH OF LIFE** Apply this code to segments of text where life expectancy, survival, length of life or its cognates are used. Includes this child or children with this disease live with this condition (live can be expressed in): • units of time (e.g. years, months) • or percentages who survive (e.g. 30% chance of surviving) • or characterisation (e.g. short-term, long-term) • and can include mention of event-free or disease-free. Excludes: • implicit statements that the child won't live (e.g. don't worry about fertility/growth/intelligence) • implicit statements that the child will live (e.g. when she grows up she will be …)
N-25	**PROGRESSION / RECURRENCE OF DISEASE**	**PROGRESSION /RECURRENCE OF DISEASE** Apply this code to segments of text dealing with progression or recurrence of the disease or pseudo-progression. Includes use of words/phrases such as: • come back • returned • happening again • got worse • metastasised • new deposits • spread • new disease
N-26	**DYING/ DEATH**	**DYING/ DEATH** Apply this code to segments of text where there is explicit use of the D word including but not limited to: • death • dying • dead • die • pass/passed away • mortality • the inevitable NB: capture all ways to say die
N-27	**SCANS**	**SCANS** Discussions about or related to all aspects of the scan (e.g. MRI, CT or PET scan). Includes discussions stemming from /part of discussion of the scan in terms of: • establishing diagnosis • what is going on with the tumour - growth, progression, stabilization, recurrence, pseudo progression • organisation of the scan • patho-physiology • tumour typing • tumour pathology/ histology • what will be done in light of the scan

[#]	Node	Definitions of node/ Instructions for Coders
N-28	**LAB WORK**	**LAB WORK** Discussions about or related to all aspects of labwork (e.g. blood tests-for purposes related to the disease or genetic counselling, CSF results) Includes discussions stemming from/ part of discussion of results in terms of pathophysiology including: • establishing diagnosis (including cancer and non-cancer) • what is going on with the tumour - growth, progression, stabilization, recurrence, pseudo progression • tumour typing • Organisation of the labwork • what will be done in light of the labwork report • All infections (e.g. meningitis, chest infection, chicken pox, thrush) • neutropenia • tumour pathology
N-29	**"OTHER" INVESTI-GATIVE PROCEDURES**	**"OTHER" INVESTIGATIVE PROCEDURES** Includes discussions of other investigative procedures and tests (e.g. audio test, cardio function, audiology, GFR, Echo, Visual, Neurocognitive, X-ray, ultrasound) that are carried out to determine a baseline and/or to assess the <u>impact of tumour Tx</u> on PT Cond. Includes discussions of: • organisation of other investigative procedures • report • what will be done in light of the report (e.g. stop chemo)
N-30	**QUALITY OF LIFE**	**QUALITY OF LIFE** Apply this code to segments of text where the words 'quality of life' or it's cognates are explicitly used or any of the following words: • quality of life/life quality • quality time • well-being/wellbeing
N-31	**SOCIAL INSTITUTIONS**	**SOCIAL INSTITUTIONS** **<u>Explicit discussions</u>** about and related to social institutions e.g. religion or school. Includes discussions about roles, responsibilities and organisation/structure: • holidays and celebrations • religious activities: Hajj, mosque, church, • make a wish type of activities (e.g. Disney and celebrity dates) • spirituality/religious beliefs and values (e.g. fasting, discussion about beliefs) • school/school attendance • home (not housing or bricks and mortar) NB. If home as a place of care is discussed, code as ACP too NB. Hospitals and hospices as social institutions appear in N-14 and N-15 OTHER SERVICES AND TEAMS WITHIN hospitals and OTHER SERVICES AND TEAMS OUTSIDE hospitals

[#]	Node	Definitions of node/ Instructions for Coders
N-32	**RELATIONSHIPS**	**RELATIONSHIPS** **Explicit discussions** about the relationships between the child, parent and HCP with others: • relationships between child (patient) and HCP • relationships between child (patient) and parents • relationships between child (patient) and their siblings • relationships between child (patient) and other family members • relationships between child (patient) and peers • relationships between parent and HCP • relationships between parent and child (patient) • relationships between parent and their other children • relationships between parent and other family members • relationships between parents and peers/friends • relationships between HCP and other family members • relationships between HCP and other HCPs • relationships between parents and other parents with a child with BTs • relationships between child and other children with BTs
N-33	**PATIENT CONDITION – PSYCHOLOGICAL STATUS & SYMPTOMS**	**PATIENT CONDITION - PSYCHOLOGICAL STATUS & SYMPTOMS** **Explicit discussions** about or related to all aspects of the patient's condition that is not physical that is not included in patient condition physical or patient and social institutions, it is restricted to psychological aspects. Includes discussions of: • character • personality • behaviour • mood e.g. angry, sad • mental health/state • coping skills/resilience/ability to deal or adapt to cancer experience NB: Does NOT include non-verbal information or observation.
N-34	**PATIENT CONDITION – PHYSICAL STATUS & SYMPTOMS**	**PATIENT CONDITION –PHYSICAL STATUS & SYMPTOMS** **Explicit discussions** about and related to all aspects of the patient's physical condition. Includes discussions of: • weight loss / gain • potential signs/symptoms of infection (e.g. wheezing, rashes, fever, red eyes) • symptoms including fatigue, loss of/disturbed consciousness, pain, nausea, breathlessness, itching, seizures • neurological function on exam (e.g. reflexes, hand-eye coordination, balance, facial palsy, swallow and speech) • allergies • cognition (e.g. memory, concentration) • daily living (e.g. dressing, eating, walking, toileting) NB: Does NOT include non-verbal information or observation. NB: Some of the discussion will stem from results of investigative procedures, including clinical exams. N.B need to be aware that neurological testing may just be reported in field notes

[#]	Node	Definitions of node/ Instructions for Coders
N-35	PARENT'S CONDITION - PHYSICAL, PSYCHOLOGICAL STATUS & SYMPTOMS	**PARENT'S CONDITION - PHYSICAL, PSYCHOLOGICAL STATUS & SYMPTOMS** **Explicit discussions** about or related to all aspects of the parent's condition, physical or psychological. Includes discussions of: • character • personality • behaviour • mood e.g. angry, sad • mental health/state • physical health/state • coping skills/resilience/ability to deal or adapt to cancer experience NB: Does NOT include non-verbal information or observation. NB: Childcare of patient to be coded under practical logistics (N-20). NB: if a parent enquires into any other treatment/medicine for their child code under this code
N-36	PARENTS PRACTICAL SOCIAL	**PARENTS PRACTICAL SOCIAL** **Explicit discussions** about or related to all aspects of the parent's practical and social responsibilities. Includes discussions of: • Housing • household chores (e.g. cooking, cleaning) • work/education/employment/visas • language • finances • childcare of siblings • pets • country of origin
N-37	OTHER FAMILY/ FRIEND CONDITION - PHYSICAL, PSYCHOLOGICAL STATUS & SYMPTOMS	**OTHER FAMILY/ FRIEND CONDITION -PHYSICAL, PSYCHOLOGICAL STATUS & SYMPTOMS** **Explicit discussions** about or related to all aspects of a family member (not parent) or friend's condition, physical or psychological. Includes discussions of: • character • personality • behaviour • mood e.g. angry, sad • mental health/state • physical health/state • coping skills/resilience/ability to deal or adapt to cancer experience NB: Childcare of patient to be coded under practical logistics (N-19). NB: Does NOT include non-verbal information or observation. NB: If another family member is mentioned and they are also a patient, code as patient physical/psychological (depending what the content is) as well as this code
N-38	OTHER FAMILY/ FRIEND PRACTICAL SOCIAL	**Explicit discussions** about or related to all aspects of a family member (not parent) or friend's practical and social responsibilities. Includes discussions of: • housing • household chores (e.g. cooking, cleaning) • work/education/employment/visas • language • finances • childcare of siblings • pets • country of origin

[#]	Node	Definitions of node/ Instructions for Coders
N-39	**HCP CONDITION - PHYSICAL, PSYCHO-SOCIAL STATUS & SYMPTOMS**	**HCP CONDITION -PHYSICAL, PSYCHO-SOCIAL STATUS & SYMPTOMS** **Explicit discussion** or mention related to any HCP's physical, psychological or social status. Includes discussions of: • character • personality • behaviour • mood e.g. angry, sad • mental health/state • physical health/state • coping skills/resilience to deal with role • cultural beliefs/language • work attendance • presentation of self and role NB: Does NOT include non-verbal information or observation. NB: This includes when they attach qualifiers to references to cases such as 'deeply upsetting' or 'sad case'
N-40	**RESEARCH PARTICIPATION**	**RESEARCH PARTICIPATION** Participation in any research other than clinical trials (e.g. HRBT project, Brightlight study). Apply this code to segments of text dealing with participation in any non-clinical trials and research participation. Including: • experiences • views of the study • relationship with researcher (doesn't include pleasantries) • seeking permission/engagement with patient/family/HCP NB: This code can be used with any research participant (e.g. healthcare professional, parent or child).

APPENDIX 6- EXAMPLE MEMO

The document below is an example of a memo that was written related alongside thematic analysis conducted in Chapter 4. These memos aided in the development and presentation of the final patterns and key differences and similarities between parents and clinicians. The below memo is on the dimension 'Construction of normality'.

29th May 2019

Different ways participant groups use normality

HCPs:

Normalise families' behaviours/feelings

Parents- help parents to not feel alone or like their reactions to this situation isn't strange.

Childs – reassure parent or child that their behaviour in this situation isn't strange and so shouldn't be worried.

Examples: Patients 12, 1, 2

Encourage normality

Normality and examples of normality are used as a tool for socialising families to the new world of having a child with a brain tumour. Many HCPs especially the palliative care HCPs and Olga encourage families to maintain normality for the child after Dx and even at the end of life. And they give examples of what they mean by normality. So at Dx- HCPs encourage continuing school in any capacity they can, encourage children to be at home where possible, continue friendships and socialising. Throughout the illness, encouraging independence of the child, attending school and allowing visitors. And even at end of life through maintaining awareness of night and day for the child.

Examples: Patients 22, 3, 6, 7

Not normal

HCPs will use the word normal when highlighting how removed from normal the child is. So ‘not normal’ or ‘we would normally see…but in X we aren’t’. it seems to be used in the case to not continue treating. E.g. if the drug was working, we would normally see… but we aren’t and so it isn’t working.

Examples: Patients 4, 7

New normal- the physical

Normality is often related to symptoms/the physical with the HCPs, and some more than others this is the new normal as HCPs will talk about certain symptoms being normal for a particular patient which of course isn’t normal as we know it but a new normal for that patient. It seems to be used in this context to normalise symptoms for the families (put them at ease) and also to act as a benchmark to signal for a need to escalate. So if a symptom is no longer what they are now ‘normally’ used to then the child may need to be brought in and further assessed.

Examples: Patients 3, 7, 8, 13, 22

Parents:

Not normal (Special)

Highlighting how their child is not normal seems to occur at different times in the illness trajectory and for different reasons. At Dx and early on in the disease progression parents state how the child hasn’t been normal for a while, how they can’t remember normal and this seems to emphasise the extreme difference the Dx and Dx journey is from the family’s previous life. Later on in the illness trajectory when the parents know how to play the game and at progression when Tx or at the very least Tx options are being withdrawn, families are either emphasising how normal the child now is (see below) or are using the fact that their child isn’t normal and is in fact special and so won’t be in the majority who don’t respond to Tx, to press the HCPs to continue Tx.

Examples: Patients 2, 22

New normal

Multiple families talk about normal in the sense of a new normal. This is related to symptoms and the physical side of things more. So talking about symptoms being normal for the child, normal drug dosing etc. something that wouldn't have previously been normal for the child or normal for any normally developing child. This acts as a communication tool between the HCPs and parents and there is an understanding that the goalposts have changed for this child and the families understand that. When not even the new normal applies anymore (not normal) this can alert the HCPs that the child may need further assessment/help.

Examples: Patients 1, 3, 7, 16

Normality as Home, school, friends- encourage HCPs to leave alone

There are many examples of what parents see as normality for their child. School mentioned a lot, home, family, friends. And families use this as a way to assert authority, let them know they know what is right and good for their child and often as a way to say leave us alone (drs and hospitals aren't normality).

Examples: Patients 6, 7, 14

Overuse of word normal to convince HCPs of health

This seems in part to allow the parents to keep treating the child. 2 families repeat throughout the encounters that 'everything is normal'. And one family even though things are far from normal, they still refer to child as 'sometimes normal'

Examples: Patients 3, 18

APPENDIX 7- NVIVO WORD SEARCH STRING

The following search string was taken from the QoL dimensions developed and used to search for all appearances of quality of life:

Normal OR normally OR normality OR normalcy OR 'everyday life' OR 'usual life' OR social OR friend OR friends OR relationship OR peer OR peers OR school OR homework OR studies OR sport OR nursery OR play scheme OR future OR control OR physical OR wish OR side effects OR late effects OR 'place of care' OR hospital OR home OR hospice OR 'spending time with the child' OR 'buying time' OR 'time left' OR sibling OR siblings OR brother OR sister OR brothers OR sisters OR family OR husband OR wife OR father OR Mother OR mum OR dad OR break OR holiday OR holidays OR Disney OR Butlins OR vacation OR trip OR weekend away OR hols OR travel OR journey OR symptom OR symptoms OR somnolence OR seizure OR swallow OR speech.

APPENDIX 8 – BLUEBOND-LANGNER ET AL (2021) ARTICLE

This is another paper to come from the wider HRBT project that I am a co-author on.

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Pediatric Blood & Cancer

PSYCHOSOCIAL AND SUPPORTIVE CARE: RESEARCH ARTICLE

Parents' responses to prognostic disclosure at diagnosis of a child with a high-risk brain tumor: Analysis of clinician-parent interactions and implications for clinical practice

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Abstract

Background: Previous studies have found that parents of children with cancer desire more prognostic information than is often given even when prognosis is poor. We explored in audio-recorded consultations the kinds of information they seek.

Methods: Ethnographic study including observation and audio recording of consultations at diagnosis. Consultations were transcribed and analyzed using an interactionist perspective including tools drawn from conversation and discourse analysis.

Results: Enrolled 21 parents and 12 clinicians in 13 cases of children diagnosed with a high-risk brain tumor (HRBT) over 20 months at a tertiary pediatric oncology center. Clinicians presented prognostic information in all cases. Through their questions, parents revealed what further information they desired. Clinicians made clear that no one could be absolutely certain what the future held for an individual child. Explicit communication about prognosis did not satisfy parents' desire for information about their own child. Parents tried to personalize prognostic information and to apply it to their own situation. Parents moved beyond prognostic information presented and drew conclusions, which could change over time. Parents who were present in the same consultations could form different views of their child's prognosis.

Abbreviations: ATRT, atypical teratoid rhabdoid tumor; DIPG, diffuse intrinsic pontine glioma; HRBT, high-risk brain tumor; MDT, multidisciplinary team meeting; UK, United Kingdom

Conclusion: Population level prognostic information left parents uncertain about their child's future. The need parents revealed was not for more such information but rather how to use the information given and how to apply it to their child in the face of such uncertainty. Further research is needed on how best to help parents deal with uncertainty and make prognostic information actionable.

KEYWORDS
cancer, communication, parent, pediatric, prognosis, prospective studies, uncertainty

1 | INTRODUCTION

Prognostic information about seriously ill children is challenging for clinicians to reveal[1] and painful for parents to receive.[1,2] Yet it is something that many argue is necessary from diagnosis forward for parents to make decisions about care and treatment for their children.[2,3] In interview and survey studies, parents report a desire for additional prognostic information.[1,4]

In order to determine what prognostic information parents were actually given and what further information they sought, we observed and audio-recorded consultations between clinicians and parents at diagnosis of their child. Analysis was directed to the content of what was said as well as the sequences of participants' statements including the responses of clinicians to parents' questions.

This is the first study to use verbatim transcripts of consultations to investigate prognostic disclosure to parents of children newly diagnosed with high-risk brain tumors (HRBT).

2 | METHODS

The data presented in this article are drawn from a larger prospective, ethnographic study of decision making for children diagnosed with HRBT at a tertiary pediatric oncology center in the United Kingdom (UK). All patients discussed at the weekly specialist neuro-oncology multidisciplinary team meeting (MDT) with diagnosis of high-grade glioma, diffuse intrinsic pontine glioma (DIPG), atypical teratoid rhabdoid tumor (ATRT), or high-risk embryonal tumor (previously high-risk CNS primitive neuroectodermal tumor) were eligible for the study.

3 | DATA ANALYSIS

3.1 | Analytic approach

Analysis was conducted from an interactionist perspective,[5] and used analytic concepts drawn from conversation analysis[6–8] and discourse analysis.[9] Analysis was confined to what was said and done in the consultation.

3.1.1 | Analysis

Consultations were observed, audio-recorded, and transcribed verbatim with nonverbal behaviors and contextual detail incorporated from embedded observers' handwritten observational notes.

Analysis was an iterative process, which began with a priori codes drawn from the literature for elements of prognosis including: Survival/Length of Life, Disease Recurrence, Cure/Curability, and Dying/Death.[1,10] Four researchers (Myra Bluebond-Langner, Richard W. Langner, Katherine Vincent, Nicolas Hall) analyzed patterns, structures, and actions grounded within the interactions using new inductively generated codes together with additional codes drawn from the literature. The final coding scheme is detailed in Figure 1.

Constant comparison was performed throughout the analytic pathway, and coding queries were run through NVivo 11 (QSR International qualitative data analysis software).

3.2 | Ethics

Per UK research guidance, data collection methods and procedures were reviewed by the Patient and Public Involvement Group for this project. All were found to be necessary and acceptable.

Advice and support were sought and received from UK Health Research Authority Confidential Advisory Group. The study was approved by the Bloomsbury Research Ethics Committee and the Research and Development departments at both sites.

4 | RESULTS

During 20 months of data collection, newly diagnosed patients and families were identified as eligible for the study at MDT. Eligible families were approached about participation sequentially until ethnographer capacity limited new enrolments. At that point, newly diagnosed cases were approached only if ethnographer resources could accommodate an additional case at the time of diagnosis. Sixteen families were approached and 13 families completed written consent to participate in the study and were followed from diagnosis forward (see Table 1 for patient, family, and household characteristics). Assent was sought

Coding Category	Codes	Type/Source	Description
1.Elements of Prognostic Information	Survival/Length of Life	A priori/Literature: Mack et al 2006, 2007	Any utterance that explicitly mentions life expectancy, survival length of life including how long child/children would/could live, survive
	Cure/Curability	A priori/Literature: Mack et al 2006, 2007	Any utterance with explicit mention of the word cure, curability/curable, incurable
	Dying/Death	A priori/Literature: Mack et al 2006, 2007	Any utterance about death or dying including words death, dying, deceased, died, passed away
	Disease Recurrence	A priori/Literature: Mack et al 2006, 2007	Any utterance about disease recurrence, progression, spread, metastases
2.Statistics and Frequencies	Statistic	Common usage	Population based statistic of cure or survival
	Natural frequency	Literature: Gigerenzer (2011)	Statistic given in the form of, e.g., 10 out of 100
	Qualification of statistic	Inductive	Any utterance mentioning replicability, up to date, or other expression implying statistic may not be accurate, e.g., a caveat
3. Non-Numeric Outcomes	Possible and most Likely	Inductive	Any scenario presented as being most likely to occur using expressions many, most, the majority, often, usually
	Possible but unlikely	Inductive	Any scenario presented as being less likely to occur using words such as few, a couple, some are an exception/exceptional, one of the ones
	Hoped for	Inductive	Any utterance characterizing outcome as hoped for, wished, preferred
	Unfortunate	Inductive	Any utterance characterizing outcome as dispreferred, unwanted
4.Features of Discourse	Framing	Literature: Tversky & Kahneman (1973)	Any utterance in which outcome is presented in terms of survival, living on the one hand or dying on the other
	Good and bad news exits	Literature: Maynard (1997)	Ending a section of the consultation with an item of "good "or "bad news"
	Proximal pairings	Literature: Leydon(2008)	Any utterance in which good news and bad news are delivered in proximity
5.Parents' responses to clinicians	Continuers	Literature: Lehtinen (2005)	Words, interjections, or gestures acknowledging what the clinician has said which allow the clinician to proceed
	Reformulations	Literature: Lehtinen (2005)	Restatement of clinicians' presentation in own words
	Questions	Literature: Lehtinen (2005)	Any question asked by a parent about prognosis
	Personalisation	Inductive	Any question or statement made by parent about the outcome for their child
6.Clinicians' response to parents' questions	Every child different/not possible to say	Inductive	Statement to the effect that population based statistics cannot be used to determine the outcome for a specific child
7. Parental Response to prognostic presentation	(Existential) Dichotomisation	Inductive	Any utterance recognizing that their child faces two mutually exclusive existential outcomes
	Existential Uncertainty	Inductive	Any utterance about not *knowing* what their child's future will be
	Recipes, Heuristics	Literature: Renjilian et al (2013), Parsons and Atkinson (2008)	Expressions used to resolve uncertainty, to make sense of the world, ease the process of assessing values or making a judgment about a course of action

FIGURE 1 Coding scheme

from children and young people as appropriate for their age and condition.

All of the 40 consultations occurring between diagnosis and initiation of treatment were analyzed. No consultations were missed. The number of consultations per case ranged from one to five (mean 3.1). Consultations were led or co-led by 12 different clinicians (see Table 1 for clinician characteristics).

Twenty-four consultations were attended by one parent, 16 by both. Additional family or friends were present at 11 consultations. Though children and young people were present during some consultations, only one adolescent made a statement about prognosis.

4.1 | Prognostic talk in the consultation

The occurrence of talk about prognosis, by either clinician or parent, was identified using four codes mentioned above. Discussion of Survival/Length of Life occurred in all 13 cases, Disease Recurrence in 11 cases, Cure/Curability in 10 cases, and Dying/Death in seven cases. Prognosis codes appeared in all 13 cases; at the first consultation in 11 cases and by the second consultation in the remaining two cases. All four codes occurred in four cases; three codes featured in seven cases; two codes in the remaining two cases. Prognosis was raised first by a clinician in eight cases, and by a parent or family member in five cases.

The four elements of prognosis talk were frequently intertwined as in Figure 2.

4.2 | Clinicians' presentation of prognostic information

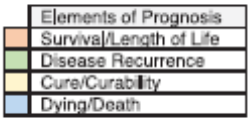
We found that clinicians presented prognosis using population-based statistics, descriptive formulations, or a combination of both.

4.2.1 | Prognosis in statistics and numbers

In seven cases, a statistic was given, in terms of population-based statistics or natural frequencies[11] (eg, 10 out of a 100). Both survival and mortality frames[12] were used to present prognostic statistics, sometimes both in immediate succession, (Figure 2, lines 11-14). The subject of the statistic varied, sometimes referring to survival at 1, 2, 3, or 5 years, at other times to cure, or to something less definite, as in "less than five patients who are doing, you know, much better."

Statistics were qualified in various ways using, for example, words and phrases such as "probably," "sort of," or "around about" (Figure 3, examples 1 and 2).

Statistics were also qualified by their presentation in conjunction with additional information. For example, in the dialogue in Figure 2 at lines 9-11, the clinician qualifies the statistic they are about to give at lines 13-14 saying that it applies to an outdated treatment modality. In other cases, clinicians quoted statistics from the treatment protocols they planned to use, allowing that figures from previous studies might not be replicated (Figure 3, examples 3 and 4).



			Commentary:
01	Clinician 6:	So that's where we are.	
02	Mother:	What are the, what are the, like, is it statistics? What —	• Mother pivots back to prognosis with a question about survival
03	Clinician 6:	Yeah —	
04	Mother:	- what —	
05	Clinician 6:	- the prognosis?	
06	Mother:	- what, what are — what's the likelihood of her recover —	
07	Clinician 6:	Okay.	
08	Mother:	- like, living?	
09	Clinician 6:	So historically — and the reason I'm saying historically is that the	• Caveat precedes presentation of statistic
10		treatment that we're using now is relatively new so obviously we're	
11		hoping it's better. But historically the treatment for children under the	
12		age of 3 with ATRT was — the survival rate at 3 years, which we take as	• Statistic is framed in terms of both survival and death
13		a surrogate, was only about 15 or 20 percent, which meant that 80	
14		percent of children probably died from their tumour at that time. The	
15		early results with the type of chemotherapy — this more aggressive	
16		chemotherapy has been better in that — but it's only — we've only really	
17		got the results over the past 2 years or so. So it seems that we're —	
18		we've made progress but of course what we're — what might happen is	
19		that those children might relapse at a later stage. So although, you	
20		know, they're actually doing better, that their tumour could come back.	
21		So I have to say at the moment we would have to say that there's still	• "Possible and most likely" formulation is paired with the "possible but unlikely" formulation
22		more of a chance that ultimately [child's name] could die of this tumour	
23		than perhaps be cured, but our cure rates have improved. So we are	
24		treating her — or I'm proposing, if you agree, to treat her with curative	
25		intent. So the idea is to tr — to cure her —	
26	Mother:	<Mother starts to cry and one of the nurses hugs her>	

FIGURE 2 Clinician's response to parent's questions about the child's prognosis (Clinician 6; Case 11: 16 months old, atypical teratoid rhabdoid tumor)

4.2.2 | Prognosis in descriptive categories

Clinicians also discussed the prognosis of children in dichotomous descriptive terms. Such talk dichotomized future outcomes. The inductive codes "possible and most likely," and "possible but unlikely," or in an alternative phrasing, "what happens to most children," and "what happens to some children" (Figure 3, examples 5, 6 and 7, 8, respectively) were developed to identify such segments in the consultations. Both formulations appeared together in four cases. The outcome for "most children" formulation appeared in six cases, and the outcome for "some children" formulation appeared in eight cases.

A second set of dichotomous categories sometimes used by clinicians in conjunction with the outcome for the most and the outcome for some was the "hoped for" outcome and the "unfortunate" outcome.

We observed no pattern to the order in which prose formulations were used when they co-occurred in clinicians' utterances. The relatively good outcome[13] was paired[14] as both antecedent and consequent to the relative bad outcome with equal frequency. For example, in Figure 2 (at lines 21-22), the "most likely" outcome ("more of a chance that ultimately [child's name] could die of this tumor") is immediately followed by the more favorable if less likely formulation that she could "perhaps be cured" (line 23).

TABLE 1 Patient, parent, household, and clinician characteristics

Patient characteristics		n = 13
Gender	Female	7
	Male	6
Age at diagnosis (years)	Median	4.8
	Range	0.9-15.7
Diagnosis	ATRT[a]	3
	DIPG[b]	2
	High-grade glioma	2
	High-risk medulloblastoma	6
Individual parent characteristics	n = 21	
Gender	Female	11
	Male	10
Coupled parent characteristics	n = 13	
Marital status	Married	11
	Other	2
English speaking	Yes	12
	No	1
Household characteristics	n = 13	
Religion	Buddhist	1
	Christian - other	2
	Muslim	6
	Not religious[c]	2
	Roman Catholic	1
	Unknown	1
Languages other than	Albanian	1
English known to be	Arabic	1
spoken in the household	Bangladeshi	1
	Mandarin	1
	Polish	1
	Somali	3
	Sylheti	1
Clinician characteristics	n = 12	
Gender	Female	7
	Male	5
Role	Consultant	10
	Registrar	2
Specialty	Oncology	10
	Neurosurgery	1
	Genetics	1

[a]Atypical teratoid rhabdoid tumor.
[b]Diffuse intrinsic pontine glioma.
[c]As stated by family.

4.3 | Parents' responses

4.3.1 | Parents' comprehension

In nine cases, parents made substantive responses[15] (ie, more than a conversational acknowledgment or continuer) to the prognostic information presented by the clinician. In the remaining four cases, parents responded during the clinician's explanations with verbal and nonverbal acknowledgments, which allowed the clinician to continue with their presentation.

Parents' responses reflected appropriate processing of the complex information they had just received by asking a concise question or making a point designed to establish that they had understood correctly. For example, at their initial consultation, the father of one child diagnosed with DIPG condensed over 50 lines of clinician dialogue—encompassing diagnosis, causation, treatment options and their efficacy, and whether the tumor is cancerous or not—into the salient point:

> "Radiotherapy, that is [you're] saying, after a period of time it [the tumor] actually comes back again?"

Parents also demonstrated understanding by reformulating points made by the clinician in their own words. For example, a mother recapitulated the clinician's reservation that the treatment protocol they would use to treat her daughter's ATRT was new and perhaps not curative, when she said:

> "No, because it hasn't been going long enough, you don't know if they, if they're cured because you're waiting to see."

4.3.2 | Parents' drive to personalize

In the majority of substantive responses parents sought to apply prognostic information to their own situation and formulate an individualized prognosis for their child.

Parents designed questions that moved from a disease population to one more closely related to their own situation. For example, the father of one child who had received published information from an American study asked:

> "How many [children treated on the protocol] in the UK?" (Case 13, Father)

Other parents sought information from a specific context, such as the specialist hospital where their child was to be treated:

> "Even at this hospital, it's never been cured?" (Case 1, Father)

Statistics with qualifiers

1. *"…the survival rates are probably somewhere between 30 and 50 percent."*

   (Clinician 12; Case 14: 15 months old, ATRT)

2. *"…we are talking sort of below 30 percent are the chances of surviving."*

   (Clinician 12; Case 5: 11 months old, ATRT)

3. *"I just think that sometimes this [published statistics of treatment protocol] might be a bit overoptimistic."*

   (Clinician 12; Case 4: 5 years old, high-risk medulloblastoma)

4. *"…we don't yet know whether we're going to replicate that [published statistics of treatment protocol]."*

   (Clinician 2; Case 13: 9 years old, high-risk medulloblastoma)

Prognosis dichotomized – "possible and most likely"

5. *"Most children do actually die from this tumor."*

   (Clinician 6; Case 1: 6 years old, DIPG)

6. *"Lots of children die from this disease."*

   (Clinician 12; Case 14: 15 months old, ATRT)

Prognosis dichotomized – "possible but unlikely"

7. *"And there are some children who've had – including at our own hospital – who've had very similar tumors, who've had chemotherapy and are alive several years later."*

   (Clinician 6; Case 12: 2 years old, high-grade glioma)

8. *"Now there is a chance that she can be cured, but that's not a huge percentage."*

   (Clinician 3; Case 9: 15 years old, high-risk medulloblastoma)

FIGURE 3 Examples of clinicians' use of statistics and prose in discussing prognosis with parents

Parents also asked clinicians about their personal experience treating children with the same diagnosis as their child:

> "I don't know if I want to ask the question, but how many survived out of your seven [the seven children you treated]?" (Case 11, Father)

Most parents explicitly asked what the outcome would be for their child (Figure 4, tiles 1, 3, 5; Figure 5, statement 6).

For some parents asking for a statistic and asking about the outcome for their child were posed sequentially as formulations of the same question. For example,

> *Mother*: What are the, what are-there's like is it statistics?
>
> *Clinician*: The prognosis.
>
> *Mother*: What, what are-what's the likelihood of her recover-her living? (Clinician 6, Case 11)

4.3.3 | Clinicians' response: uncertainty

Clinicians frequently responded to parents' questions by invoking the uncertainty of individual-level predictions (Figure 4, tiles 2, 4, and 6). They frequently cautioned that each child is a unique individual; further, prognosis would be contingent upon the response to treatment:

> "So I think everyone's very, very different and we will need to see what kind of response she has to the radiotherapy." (Clinician 4, Case 1)
>
> "It's, it's quite difficult because it depends on whether or not he responds to the chemotherapy." (Clinician 6, Case 12)

Clinicians acknowledged that they simply did not know what the outcome would be:

> "Well I think we have to be a little bit guarded about that because the honest answer is we don't know." (Clinician 5, Case 7)

The message to parents, implicitly and explicitly, was to wait and see. As one clinician put it

> "I think that's all that we can do." (Clinician 4, Case 1)

4.3.4 | Parents' persistence

Some parents persisted in their search for personalized information. In Figure 4 for example, the mother's pursuit of a personalized prognosis is evident. She responds to the clinicians' statements of uncertainty by reformulating the same question about her son's chance of survival on three separate occasions over three consecutive consultations led by two different clinicians. Each iteration of her question is met with the same response from the clinicians:

> "it's difficult" to predict."

4.4 | Parents' application of the clinician's message

In case 1, three findings emerged about parental response to clinicians' prognostic disclosure. First, the parents exhibited the use of what has been termed explicit heuristics or recipes[16,17] (Figure 5, consultation 3). These were used to resolve their uncertainty about their child's future as either "buying time" (mother) or as "after if it all goes well… it could be something very, very good" (father).

Second, the sum of prognostic disclosure did not lead to a single unique application of it to that child, concerning that child's future (Figure 5, consultation 3). Two parents present throughout the diagnostic consultations applied the same information from the clinician differently.

Third, a parent's application changed over time. In this same case while the parents were aligned in the first two consultations, both envisioning the "most likely," "unfortunate," outcome that their daughter's condition is incurable (Figure 5; statements 1-4), by the third consultation they were no longer aligned; the mother's interpretation having remained unchanged during the three consultations. In Figure 5, we see the father's interpretation evolve over 3 days. He moves from a view of his daughter's condition as incurable, to a view that the outcome is not necessarily predictable (Figure 5, statement 5), to a formulation of the unlikely outcome that his daughter could be the exception (Figure 5, statement 7).

5 | DISCUSSION

The goal of this study was to observe in situ what prognostic information was actually given to parents, how it was presented, and parents' voiced responses to the information presented, particularly what else parents wanted to know about their child's prognosis at the time of diagnosis.

We found clinicians' behavior in line with guidance[18] to communicated directly and honestly with parents. Parents of children with HRBTs were told that the majority of children with such tumors do not survive. At the same time, highly unlikely outcomes were not dismissed as impossible or as unrealistic. Clinicians expressly stated that the odds,

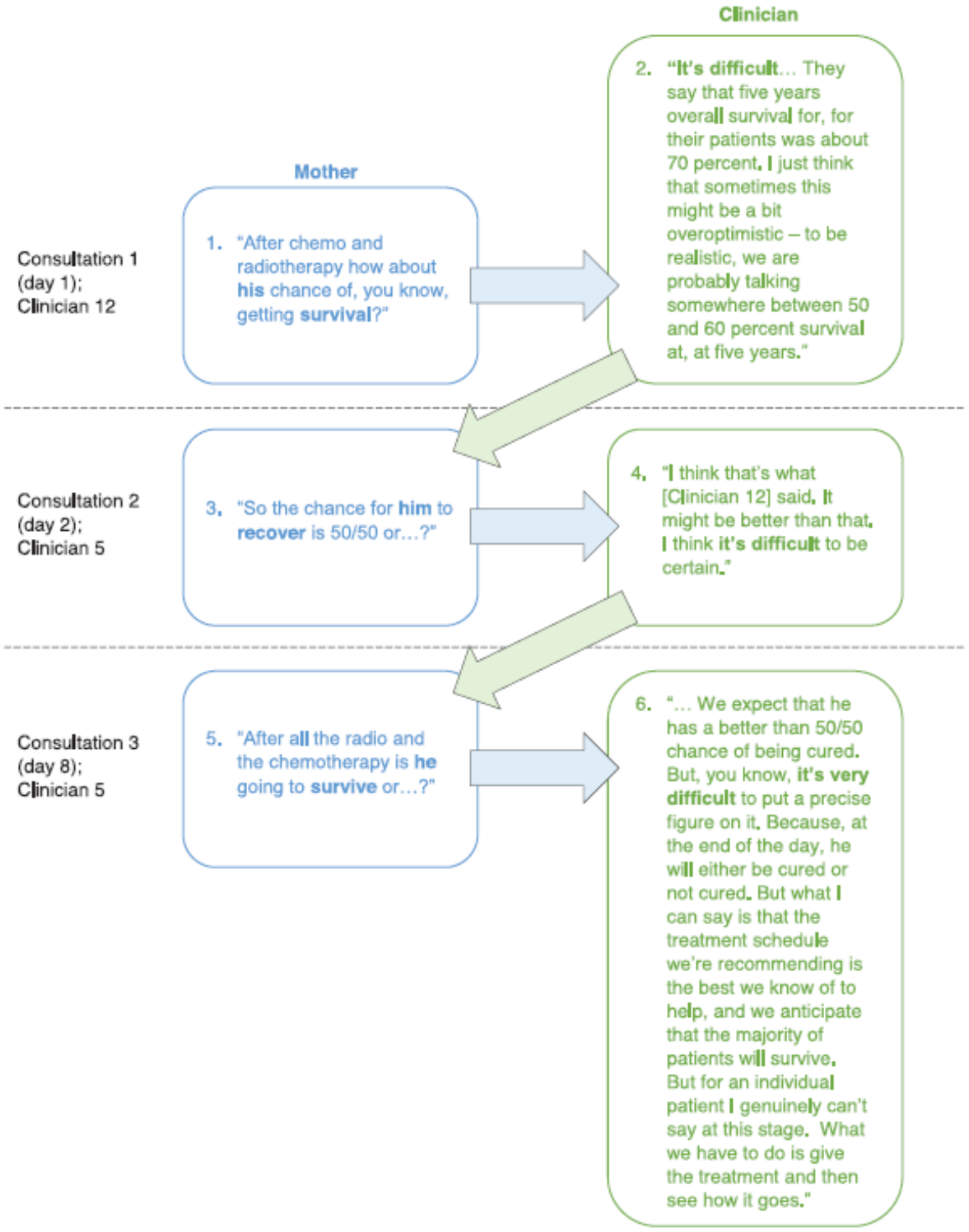


FIGURE 4 Common pattern in discussion of prognosis: Parents asking for more patient-specific information, clinician responding with more general statement of prognosis (Clinician 12 and Clinician 5; Case 4: 5 years old, high-risk medulloblastoma)

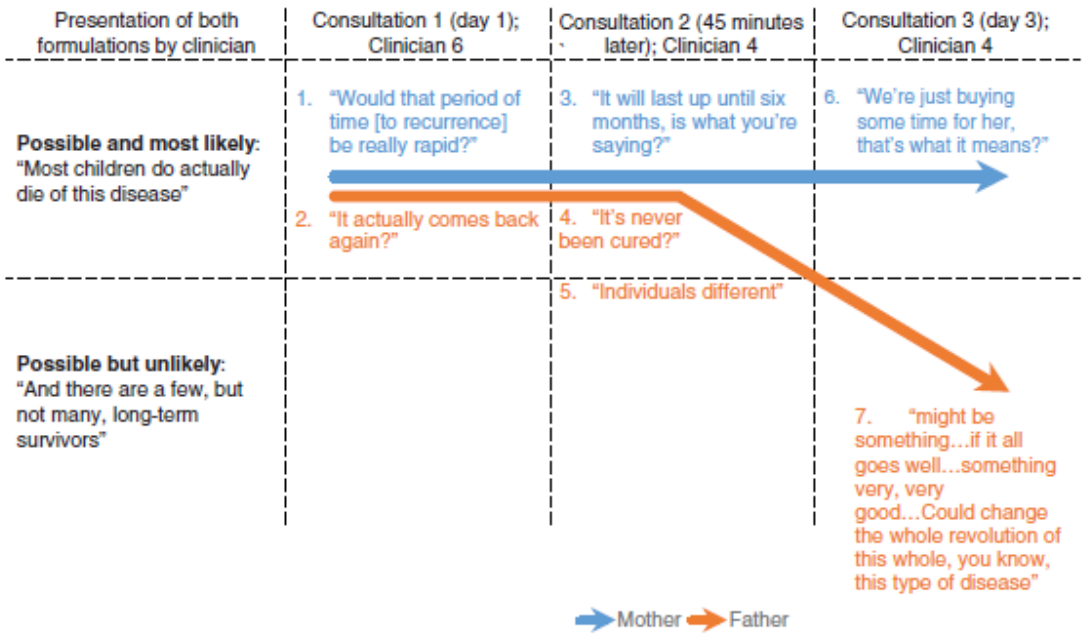


FIGURE 5 Differences in parents' responses when presented with the same formulations of the prognosis (Clinician 6 and Clinician 4; Case 1: 6 years old, diffuse intrinsic pontine glioma)

however low, were not zero. Clinicians provided statistics in more than half the cases, but also usually qualified these statistics. None of the clinicians prioritized statistical understanding. They just as often used descriptive categories in talking about the child's prognosis.

The receipt of a poor prognosis diagnosis puts parents in the position of having to redefine their child's and family's future.[19,20] Our findings suggest that prognostic information, including population-level statistics, was not by itself sufficient for parents to use to accomplish this. We observed parents in the consultations moving beyond the information given them to construct a personally meaningful and actionable account.

Through their questions about prognosis, parents searched for personalized information rather than population-based information. They asked about outcomes in the hospital where their child was being treated, about the outcomes of past patients of the treating clinician, and about what was going to happen to their child.

Parents used a binary framework to frame prognostic information. Would their child survive or die? Would it be 0% or 100% for them? Here the numbers are metaphors for something nonnumeric and existential.

Parents focused on a unique, unrepeatable event. Their question was one for which no probabilistic or frequency data about similar events were relevant. As a result, they were left with an unresolvable uncertainty.

The case in Figure 5 illustrates two parents dealing with uncertainty about their child's outcome. They each came to different resolutions between the most likely outcome, that their child would die, and the unlikely though possible outcome, that their child would survive. They used terse characterizations of the situation: "just buying time" and "a revolution."

Past research in other cancer populations and in other diseases has found parallels to what we observed in these parents of children with HRBTs. Renjilian et al[16] found that parents of children with life-threatening illnesses faced "an irreducible amount of uncertainty."[16(e567)] In such a situation, parents used "explicit heuristics"[16] in the form of aphorisms, mantras, or rules of thumb. These heuristics are distinct from the implicit heuristics of behavioral psychologists[12] in that their goal is not to estimate probability but to "help parents to make sense of the world, ease the process of assessing values and [cast] judgment about a course of action."[16(e567)]

Such heuristics were used by parents to frame the problem, and to imagine and commit to courses of action. In Figure 5, we see such heuristics being deployed. The mother sums up her child's condition and treatment as "just buying time." The father embraces the possible but unlikely trajectory and rests the child's future in the hands of progress in treatment. Each of these is what Renjilian et al[16] describe as an explicit heuristic.

The response of parents of children with HRBTs also parallels what has been found in studies of parents receiving genetic counseling. Recipients of statistics about carrier and reproductive risks translated this information into binary descriptive categories and found themselves in a state of uncertainty about the outcome for their child. They personalized statistical information and made it usable by developing "recipes," which made information actionable rather than paralyzing.[17]

Given the question that parents want to answer, their uncertainty could not be resolved by more robust knowledge or understanding of

the probability or frequency of similar events. Renjilian et al[16] suggest that the heuristics parents elect to use may be linked to what they perceive as required by their role as a parent. The situation in which parents find themselves at diagnosis is that of defining their identity as the parent of a child with cancer. We suggest that parents' interpretation of prognostic information may be part of establishing their changed identity and that of their child. Prognostic interpretation is thus an act that engages parents as both social and moral actors.

It is essential that these parental acts be better understood, because it may be that in transforming statistical information into descriptive, binary categories and personalizing the clinician's prognostic information and making it actionable,[16,17] we may have arrived at an empirically developed account of parents' prognostic understanding, which stands between the clinician's communication and parent's subsequent decisions about their child's care and treatment.

While the sample is small, it is consistent with prevalence and expected referral patterns as well as representative of the HRBT population in the UK where each year approximately 100 children are diagnosed with HRBT. The 16 patients identified by the MDT represent roughly 10% of the total number of newly diagnosed HRBT patients in the UK during the 20-month period. The religious and linguistic diversity of the sample reflects the wider population of patients diagnosed with HRBT in the UK and their families. The sample included parental dyads and was gender balanced: fathers and mothers were recruited and participated.[21]

A primary role of qualitative research is to uncover process, rather than to generate statistically significant findings about populations.[22,23] A number of factors contribute to the quality of the analysis presented here. The data have been collected using what Kaye et al[24] have termed a gold-standard methodology for the investigation of clinician communication. The method of real-time recording eliminates issues of recall bias and insures the accuracy of our accounts of what participants said and did.

Established theories[19,20] and previous research in pediatric cancer and other areas support our analysis of parents' response to prognostic information in that these responses derive from fundamental patterns of response to serious illness. These theories and findings add credibility to our finding that parents facing an irresolvable existential uncertainty about their child's future may resolve this situation through explicit heuristics or recipes that parents employ to define a future for themselves and their child and to repair the disruption of diagnosis of a poor prognosis illness.

Further research is needed to extend these findings to other pediatric cancer and serious illness populations. This would include a search for negative and deviant cases[25] in which for example, in an effort to come to terms with their uncertainty about prognosis, the parent sought more population-based statistics. Through the discovery of such cases we would learn of limits to or conditions which might be placed on the findings presented here.

The aspects of parents' appreciation of prognostic information, which this study has pointed up have implications for the important question of what constitutes optimal clinician communication about prognosis with parents and how to achieve it. There is a critical element of parents' response to prognostic information, which does not fit within the straightforward model of the delivery and uptake of a message of factual information. This additional element is not simply the parents' emotional reaction to prognostic information. It adds social and ethical dimensions as well.

From a clinical perspective, the study also demonstrates the importance of listening to parents' formulations and applications of the information they receive. Recipient's form interpretations of what is said regardless of the clarity of the "message." This independent dimension of the communication process needs further attention in communication research. An exclusive focus on the clinician's use of language and nonverbal gestures in solving perceived shortcomings in clinician-parent communication addresses only one side of the process.

Evidence of parental interpretation and realization can be found within the consultation itself. Learning to recognize these parental contributions is vitally important.[26] Clinicians can on the basis of this understanding better detect parents' specific needs and goals, and engage parents accordingly.

Clinician communication training must therefore address ways of sensitively eliciting parents' points of view. Direct questions may not be the most effective way of helping parents to display their understanding,[27,28] especially at diagnosis. Further research is needed to provide clinicians with resources to facilitate parents' expressing their understanding of their child's condition and prognosis, and what further information they seek.

This study also points up that parents of children with poor prognoses experience fundamental uncertainty at diagnosis. Managing uncertainty, Snaman et al[29] wrote, is an essential part of patient-centered care. Families would benefit from research into how this intractable uncertainty affects daily life and into how adverse effects can be mitigated.

In summary, what is needed to improve clinician-parent communication lies not just in furthering the parent's understanding of the clinician, but in developing the clinician's understanding of the parent as well.

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CONFLICT OF INTEREST

The authors declare that there is no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

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