

**Coping with a childhood brain tumour:
A qualitative analysis of parents' experiences**

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THESIS DECLARATION FORM

I 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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OVERVIEW

Pre-existing research on the stress reactions of caregivers of children with brain tumours was reviewed. Four overarching stress reactions were notably present for parents: burden from adjusting to changes in routine, burnout from fatigue and emotional exhaustion, residual stress from diagnosis and treatment, and future-oriented uncertainty. There is evidence to suggest that psychosocial implications for parents are a concern and that they require support from professionals long into the survival period.

As part of the empirical research, ten parents of paediatric brain tumour survivors were retrospectively interviewed about their experiences of coping from diagnosis through to the survival period. Interviews were transcribed and four domains were devised from a thematic analysis: *Focusing on the here-and-now* in which parents concerned themselves with taking one day at a time rather than thinking about what may arise later; *Overcoming helplessness* reflected the desire to provide care-giving duties; *Different needs met across the system* included emotional bonding with other parents on the ward, whilst wanting family to offer respite; *Finding a new normal* featured in the survival period when parents reflected on new values for the family. Coping mechanisms were seen as a process, changing dependant on the time period.

The literature review and empirical study are rounded off by a critical appraisal of the research process, which focuses on the clinical utility of working qualitatively with a paediatric brain tumour population, a discussion of homogeneity versus heterogeneity when sampling, and an appraisal of thematic analysis.

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Part 1: Review Paper

Parental stress reactions in paediatric brain tumours:

A narrative synthesis

ABSTRACT

Aims: This review explores the association between parental stress reactions and paediatric neuro-oncology, together with a methodological critique of the included studies.

Methods: MEDLINE, PsycINFO and PubMed were used to source studies, both quantitative and qualitative in design, investigating parental stress reactions to childhood brain tumours.

Results: Fourteen studies were identified. Despite broad methodological designs and varying conceptualisations of stress, there was general accord across studies that stress is clinically debilitating for many parents. Synthesis indicated four key stress reactions: burden from adjustment to the caregiver role, burnout from physical fatigue and emotional exhaustion, residual stress from time of diagnosis and treatment, and future-oriented uncertainty.

Conclusions: Lack of study comparability and small samples are problematic but there is evidence that parental stress reactions are a common consequence of childhood brain cancer, continuing post-treatment and often left unattended. Recommendations are given for clearer detection through standardised assessment.

INTRODUCTION

Survivors of childhood brain tumours are an increasingly important population to study due to improvements in treatment outcomes and subsequent lower mortality rates, with over 65% of children living for over five years after diagnosis (McKinney, 2004; McKinney, 2005). With more rigorous treatment types and potential tumour recurrence, much focus has been placed on the child's psychosocial and neurocognitive functioning, with statistics highlighting difficulties for the majority of survivors. For example, approximately 60% of children will be left with significant difficulties, including physical, intellectual, behavioural, social and temperament impairments (Anderson, 2003; Carpentieri, Waber, & Pomeroy, 2003; Patenaude & Kupst, 2005; Upton & Eiser, 2006). However, research has chiefly focused on the patient alone. Whether the child survives or enters palliative care, there is a paucity of research on caregiver mental health and the ability to cope with caring for a recovering, or recovered, child.

Childhood cancer is not only distressing to the patient but to the parent, family and wider system, with early research conceptualising cancer as a family disease (Chesler & Barbarin, 1987; Binger et al., 1989). A commonly-held definition of stress is that it occurs when the demands of a task exceed the resources a person has to manage them (Lazarus, 1966); therefore, the consequences of parents facing stress can be debilitating to a child's care in which the parent is unable to meet the roles required as both nurturer and medical caregiver. Findings of elevated parenting stress have been shown across paediatric medical settings, for example, with traumatic brain injury (Hawley et al., 2003; Verhaeghe, Defloor, & Grypdonck, 2005), learning disabilities (Hassall, Rose, &

McDonald, 2005), and developmental disorders (Lopez, Clifford, & Minnes, 2008; Mori, Ujiie, & Smith, 2009).

The new role of carer as well as parent can be burdensome, tiring and emotionally-charged (Van Hooft, 2010). Parents must bear the burden of caring for a child under distressing circumstances as well as their own stress reactions, which include psychological and physical reactions in response to adapting to new conditions that may accompany these changes in adjustment. These often require more time and effort than prior to the illness-onset. Furthermore, complicated and exhausting treatment regimes, including medication adherence and attending appointments, as well as potential deterioration related to illness progression or treatment effects, can increase the demands of the caregiver, who may find their new role distressing (Beigel, Sales, & Schulz, 1991).

Stressors, such as burnout and burden, can be explained within the Transactional Model of Stress and Coping (Lazarus & Folkman, 1984), in which the parent may not be able to manage the number of internal and external pressures put upon them. Similarly, Patterson's Family Adaptation and Adjustment Response Model (1988) implies that the parent's capabilities, such as coping tools and resources, may be outweighed by cognitive and behavioural demands.

Current Reviews of the Literature

A recent systematic review by Vrijmoet-Wiersma et al. (2008) explored parental stress reactions for all childhood cancer types within sixty-seven studies between 1997 and 2007. The researchers found that stress was especially prevalent around time of diagnosis, higher for mothers than fathers, and that parents were on the whole resilient

with the exception of a subset who continued to present with stress. In particular, they found four main subsets of stress: the pervasive uncertainty of relapse, the anxiety and apprehension of their child's wellbeing, depressive feelings of both hopelessness and helplessness at diagnostic levels, and a high proportion of parents facing moderate to severe levels of post-traumatic stress. These areas of stress were reported to have clinically significant overlap with one another, with suggestions that earlier management and interventions could be beneficial.

Other research validates these findings. Investigated stress reactions typically concern feelings of helplessness, uncertainty, low control and experiencing the diagnosis and treatment as akin to a trauma, even long after the trauma itself (Barakat, 1997; Grootenhuis & Last, 1997). Even during the survival period, which is typically given as any time up to three or, in some studies, five years post-treatment, stress can manifest itself within adjustment to new family setups and daily difficulties, including physical, financial and social stressors, and can result in parents feeling unable to cope (Van Dongen-Melman, Zuuren, & Verhulst, 1998).

Existing research has focused on parental stress reactions within all presentations of childhood cancer, often grouping together brain tumours with other types of cancer, and finding childhood cancer to be a period of chronic stress for parents (Hoekstra-Weebers et al., 2001; Barrera et al., 2004). However, some literature suggests that brain cancer may affect parents differently, due to their awareness of the neurocognitive sequelae and potential restrictions on their child's life (Radcliffe et al., 1996). Sherwood and colleagues (2004) argue that the diverse multitude of symptoms that stem from brain cancer is enough to warrant this illness an entirely different disease compared to other

cancers. Due to central nervous system location and potential neurocognitive compromise, as well as on-going late effects of treatment and the extension of the caregiver role for many years to come, possibly even indefinitely, neuro-oncology can be seen as a different medical condition to other types of cancer (Ressler, Cash, & McNeill, 2007). As such, carers of children with this condition should be seen as a separate population; one that has a lack of targeted data available. Findings support this, such as higher levels of post-traumatic stress identified in parents of brain cancer patients compared to other cancers (Manne, DuHamel, & Redd, 2000).

Aims

Research indicates that parental stress is significant for childhood cancer and that brain tumours can have distinct outcomes compared to other cancers. However, there are no reviews that incorporate research carried out with parents regarding stress reactions from brain tumours alone. Therefore, a narrative synthesis was employed to review and evaluate research of parental stress reactions within the parameter of childhood brain cancer alone, to determine whether these stress reactions are similar to other cancers or whether different procedures should be put in place by healthcare professionals when considering the support parents may require.

METHOD

Criteria for considering studies

Types of studies

In order to be eligible for this review, all studies had to explore factors relating to parental stress, regardless of control/comparison group or type of design. The focus could be any as long as it featured at least one aspect of stress either through questionnaires measuring levels of stress quantitatively or by researchers qualitatively asking parents to consider stress.

Any aspect of stress was eligible, including but not limited to stress reactions previously believed to be implicated, such as psychological stress, post-traumatic stress, burnout, and stress-related anxiety. Studies looking purely at interventions for stress and/or those who did not rate levels of parental stress were excluded.

Only studies investigating brain tumours, or those in which brain tumour data could be distinguished and separated out from other cancers, were included.

Types of participants

Participants were stated to be any parent or primary caregiver of a child with a brain tumour diagnosed and treated in childhood up to the age of eighteen years. No discrepancy was given for different brain tumour types or treatment types due to small sample sizes, although a limitation is that these can lead to different survival and relapse rates as well as varying neurocognitive and other disabling sequelae, which in turn can potentially affect the caregiver's stress reactions.

Types of measures

A preliminary search detected that some of the published research was qualitative and contained valuable information about parental stress. Therefore, both quantitative

and qualitative studies were included in this review. Studies must have reported a quantitative measure of stress or qualitative themes related to parental stress drawn from transcriptions of semi-structured interviews, conducted by the researchers either with parents individually or within focus groups.

Search methods for identification of studies

Electronic searches

Searches were run in MEDLINE, PsycINFO and PubMed in June 2013 with no parameters given to year of publication. The search used keywords (parent OR caregiver) AND (brain cancer OR brain tumour OR neuro-oncology) AND stress.

Only peer-reviewed published articles were included. Studies investigating all kinds of childhood cancer were read to determine if specific results for brain cancer could be distinguished.

Reference lists of each study included in the review were manually searched to identify possible other sources.

Data collection and analysis

Selection of studies

Initial searches identified 1,024 citations. Titles and abstracts were examined against the listed inclusion and exclusion criteria, with many studies focusing solely on medical

trials, without psychosocial outcomes, or without distinction between brain tumours and other childhood cancers.

Seventy-six citations could not be excluded and, of these, full texts were sourced for inclusion and exclusion criteria. A further 26 articles were sourced from reference lists but none met the inclusion criteria after reading the abstracts. See Figure 1 for screening and eligibility flow.

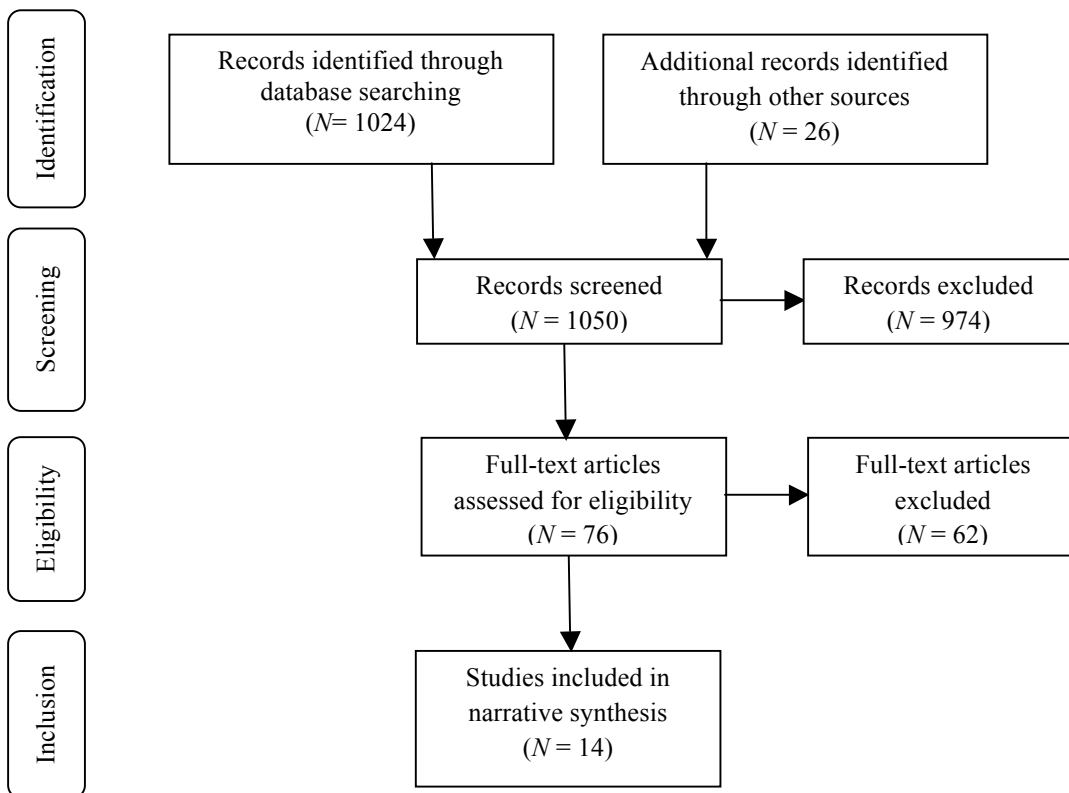


Figure 1. Information flow on study selection and inclusion

Data extraction and management

Data were extracted, covering demographic information such as parent gender, time since diagnosis, and details of the stress reactions being addressed. For quantitative

studies, measures and outcomes were collated. Findings from qualitative studies were entered thematically. Methodological designs were assessed according to a number of criteria adapted from guidelines by Letts et al. (2007) for qualitative studies, and from the Downs and Black checklist (1998) for quantitative studies (see Appendices A-B).

Data synthesis

Given the data were both quantitative and qualitative, and different measures were used within the quantitative studies thus making statistical comparison difficult, a narrative synthesis approach was used. This methodology allows for systematic and transparent comparison and evaluation (Popay et al., 2005) and the findings are derived with the view of informing guidelines for clinical practice (Arai et al., 2007). Narrative synthesis can be beneficial for reviews in which statistical analyses are not appropriate, especially when qualitative methodologies are also being examined.

Narrative synthesis follows a framework of tools and techniques to allow for robust conclusions. Within this synthesis, textual descriptions of each included study were outlined to identify the outcomes and a preliminary synthesis of areas of parental stressors was developed. Following this, themes within qualitative data were transformed to meet a common rubric and findings from quantitative studies were tabulated, and conceptually mapped onto an 'idea-web', which allowed for exploring the relationships between the studies as a visual representation of the common themes (Popay et al., 2005). Findings were clustered and vote counting determined the overarching domains of parental stressors.

RESULTS

Description of studies

Fourteen studies met the inclusion, with four being qualitative and ten quantitative. All studies considered stress reactions for both mothers and fathers, with the exception of one study that focused solely on fathers (Bonner et al., 2007) and one on mothers (Shortman et al., 2012). Notwithstanding, the majority of respondents in the studies were female and some were comprised of all mothers (Wideheim et al., 2002; Keir, 2007). Most of the studies focused on a limited number of stress reactions, such as physical and emotional burnout, and few explored their relationship to other demographic factors, including marital status. The majority of studies used convenience samples with limited comparison to controls. No study was longitudinal and few compared different illness phases. See Table 1 for characteristics of included studies.

Table 1. Characteristics of included studies

Study	Demographics	Measure(s) ¹	Results
Bennett et al. (2013)	<i>N</i> 37 Age (Mean) 39 Gender (%F) 87 Status (%Couple) ² 76 Ethnicity (%White) 84 Country UK Illness Phase ³ All	PLOC, PSI/SF, WAYS	51% experience clinical stress; self-blame and external locus of control elevate stress
Bonner et al. (2007)	<i>N</i> 46 Age (Mean) 40 Gender (%F) 50 Status (%Couple) 78 Ethnicity (%White) 87 Country USA Illness Phase All	BSI, CGSQ, IES, IFS, PEGI	No difference in stress levels between genders; majority present clinical stress levels
Bruce et al. (2010)	<i>N</i> 52 Age (Mean) 42 Gender (%F) 88 Status (%Couple) Unknown Ethnicity (%White) Unknown Country UK Illness Phase Post	IES-R, PACHIQ-R-P	29% experienced clinical post-traumatic stress symptoms, only 1 parent experienced no symptoms; poor conflict resolution and more tumour reoccurrence predicts stress
Freeman et al. (2004)	<i>N</i> 139 Age (Mean) 43 Gender (%F) 56 Status (%Couple) 84 Ethnicity (%White) Unknown Country USA Illness Phase All	Validated survey	Different stressors at each phase, including unmet information need and child's emotional changes; marriage resulted in higher stress
Fuemmeler et al. (2001)	<i>N</i> 28 Age (Mean) 42 Gender (%F) 64 Status (%Couple) 86 Ethnicity (%White) 86 Country USA Illness Phase Post	PDS, PPUS, WAYS	42% meet criteria for PTSD; associated with emotion-focused coping and perceived uncertainty; time since diagnosis and gender not factors
Hutchinson et al. (2009)	<i>N</i> 90 Age (Mean) 39 Gender (%F) 81 Status (%Couple) Unknown Ethnicity (%White) 87 Country USA Illness Phase All	CGSQ, IES, IFS, PEGI	Lower distress post-treatment but continued uncertainty and burden

¹ Measures: BSI = Brief Symptom Inventory; CGSQ = Caregiver Strain Questionnaire; CSI = Caregiver Strain Index; DASS = Depressive Symptoms Anxiety Stress Scales; IES = Impact of Event Scale; IES-R = Impact of Event Scale – Revised; IFS = Impact on Family Scale; PEGI = Parent Experience of Child Illness; PLOC = Parental Locus of Control Scale; PACHIQ-R-P = Parent-Child Interaction Questionnaire - Revised Parent Version; PDS = Post-traumatic Stress Diagnostic Scale; PPUS = Parent's Perception Uncertainty in Illness Scale; PSI/SF = Parenting Stress Index – Short Form; PSS = Perceived Stress Scale; SMBQ = Shirom-Melamed Burnout Questionnaire; WAYS = Ways of Coping

² Status (%Couple): Percentage of those in a relationship, whether married or not

³ Illness Phase: All = Combination of Treatment and Post-Treatment; Post = Post-Treatment

Table 1. Continued

Study	Demographics		Measure(s)	Results
Keir (2007)	<i>N</i>	5	PSS	All presented elevated stress; demand for stress reduction techniques
	Age (Mean)	48		
	Gender (%F)	100		
	Status (%Couple)	100		
	Ethnicity (%White)	100		
	Country	USA		
	Illness Phase	Post		
Norberg (2007)	<i>N</i>	44	SMBQ	Burnout associated with emotional exhaustion and cognitive difficulties but not time since treatment; higher burnout for mothers
	Age (Mean)	41		
	Gender (%F)	55		
	Status (%Couple)	100		
	Ethnicity (%White)	Unknown		
	Country	Sweden		
	Illness Phase	Post		
Norberg and Green (2007)	<i>N</i>	4	Thematic Analysis	Stress from daily stressors, including adjustment, exhaustion, threat of relapse, neurocognitive sequelae
	Age (Mean)	Unknown		
	Gender (%F)	50		
	Status (%Couple)	100		
	Ethnicity (%White)	100		
	Country	Sweden		
	Illness Phase	Post		
Norberg (2009)	<i>N</i>	11	Thematic Analysis	Stress related to uncertainty about child's wellbeing and future, physical and psychological exhaustion
	Age (Mean)	Unknown		
	Gender (%F)	64		
	Status (%Couple)	Unknown		
	Ethnicity (%White)	Unknown		
	Country	Sweden		
	Illness Phase	Post		
Norberg (2010)	<i>N</i>	44	PSS, SMBQ	Stress not related to work or time since treatment, but associated with tangible stressors and existing challenges
	Age (Mean)	41		
	Gender (%F)	55		
	Status (%Couple)	100		
	Ethnicity (%White)	Unknown		
	Country	Sweden		
	Illness Phase	All		
Ownsworth et al. (2009)	<i>N</i>	27	CSI, DASS	Stress associated with depressive symptoms and strain of the caregiver role
	Age (Mean)	57		
	Gender (%F)	44		
	Status (%Couple)	Unknown		
	Ethnicity (%White)	Unknown		
	Country	Australia		
	Illness Phase	Post		
Shortman et al. (2012)	<i>N</i>	6	Thematic Analysis	Stress associated with uncertainty of future, disempowerment during diagnosis, functional adjustment, and parenting style conflict
	Age (Mean)	39		
	Gender (%F)	100		
	Status (%Couple)	83		
	Ethnicity (%White)	Unknown		
	Country	UK		
	Illness Phase	Post		

Table 1. Continued

Study	Demographics		Measure(s)	Results
Wideheim et al. (2002)	<i>N</i>	3	Content Analysis	Caregiver burden, uncertainty, helplessness, and receiving negative information lead to stress
	Age (Mean)	42		
	Gender (%F)	100		
	Status (%Couple)	Unknown		
	Ethnicity (%White)	Unknown		
	Country	Sweden		
	Illness Phase	Post		

Overall stress

A substantial proportion of parents, whether at treatment or post-treatment, present with levels of clinically significant stress. For example, Bennett et al. (2013) found 51% of parents have clinically elevated stress, and Bruce et al. (2010) found 29% present with severe levels of post-traumatic symptoms to name but two of the quantitative studies. All of the participants in the qualitative studies spoke of feeling overwhelmed and burdened with the role of caregiver.

Stress can manifest itself in a number of ways, and is related to the diagnosis itself, taking on the role of caregiver and the exhaustive duties it calls for, as well as feeling the psychological distress of unprocessed emotions. No study found that this population was akin to the general population for levels of stress. All studies suggest that stress reactions are a serious enough issue to warrant investigation into effective interventions.

There is considerable overlap between parental stress reactions for paediatric brain tumours and all cancers, as seen in the larger systematic review (Vrijmoet-Wiersma et al., 2008) but there are some noticeable differences. A more elevated fear of relapse and future neurocognitive deterioration is one aspect that is specific to brain tumours, possibly due to the central nervous system location (Ressler, Cash, & McNeill, 2007).

Secondly, post-traumatic stress is rated higher in the brain tumour group, possibly due to the higher likelihood of mortality.

The majority of the studies demonstrated analogous results, with the exception of some elements relating to contradictory stress reactions that are discussed in the refutational factors section.

After synthesising the data, what follows are four sections, each relating to a different type of stress reaction, although it must be remembered that there is overlap between these areas and that causal relationships are difficult to define, since vicious cycles between any two or more of these factors may result in further inability for the caregiver to cope.

Burden from adjustment to the caregiver role

Parents are expected to take up the extra role of caring for a sick child so that, on top of daily tasks, such as taking their child to and from school, and providing nurture and support, there are also the added activities of giving medication, taking the child to hospital appointments and any new adjustments to the child's role, such as managing a wheelchair (Wideheim et al., 2002; Norberg & Green, 2007; Ownsworth et al., 2009; Norberg, 2010; Shortman et al., 2012). Whilst this can lead to emotional and physical exhaustion, I separated this by considering the adjustment to the caregiver role as an interpersonal stressor, and the burnout from exhaustion as an intrapersonal stressor, and consequently discuss those features in the following section.

Adjusting to the caregiver role can result in stress during treatment and post-treatment, as the caregiver role takes on differing functions related to daily stressors. For

example, at treatment, hospital visits and caring for a sick child can be stressful whereas the functional adjustment due to changes in the child's mobility and neurocognitive changes will affect previously formed routines. One of the few studies to investigate more than one illness phase found that, at time of treatment and three and six months after that, being readily available to the patient in providing support was detrimental to the parent's own health (Wideheim et al., 2002). Further, this support limited parents' ability to maintain other aspects of life, such as household duties, keeping in touch with friends and losing sleep due to being busy with their duties.

Additionally, the caregiver role can throw up new situations the family has not faced before, and the parent may not have the coping mechanisms or strategies to resolve these problems. Stress reactions can come from two sources of conflict, either the inability to resolve conflict with the child, especially when feeling guilty about setting boundaries or disciplining a sick child, as well as conflict between two parents who have markedly different ways to parent a sick child, again, something that may not have been present in their relationship prior to the diagnosis of cancer (Bruce et al., 2010; Shortman et al., 2012).

One study verified that the caregiver role in particular can lead to stress compared to stresses from other aspects of the parent's life by controlling for employment pressures (Norberg, 2010). This study asked parents to rate how demanding, stressful and manageable their work was, and determined that there was no association with parental stress. Norberg also hypothesises that employed caregivers may either lower their work demands or take a more relaxed attitude to work so they can focus on their child instead.

Burnout

Although the burden of the caregiver role comes from the extra tasks of caring for one's child, thus a relational and dynamic function of care, this can also lead to the solitary reaction of burnout, the physical and emotional exhaustion from an overload of responsibility. Burnout can also cause and be caused, in a vicious cycle, by depressive symptoms. Seven of the fourteen studies examined at least one aspect of burnout, which can be further broken down into physical fatigue, emotional exhaustion, and depressive symptoms (Wideheim et al., 2002; Keir, 2007; Norberg, 2007; Norberg & Green, 2007; Norberg & Steneby, 2009; Ownsworth et al., 2009; Norberg, 2010).

Physical fatigue overlaps with adjustment to the caregiver role, in which the parent may be expected to complete daily tasks with the additional duties of attending hospital appointments and physically caring for the child. Norberg and Steneby (2009) found that parents were concerned their children would be missing out on academic input and cognitive stimulation and, with feelings of inadequacy as substitute teachers, parents spend much time helping them with homework. Extra time and resources that needed to be included in the daily routine could also lead to burnout, such as preparing special meals to aid a child with poor digestion and appetite. Overprotection, due to the view that the child was fragile, led to a more physically demanding workload, such as carrying the child when parents believed he or she was not physically strong enough to walk (Norberg & Steneby, 2009). Norberg (2007, 2010) found that time elapsed since end of treatment did not affect levels of exhaustion, although sample sizes were small.

Emotional exhaustion occurred for parents who were preoccupied with taking on new roles, for example, support worker, teacher, medical assistant and employing

psychological tasks, including supporting the worried and depressed child (Wideheim et al., 2002; Norberg & Green, 2007). Any neurocognitive changes, such as difficulties with attention, language and memory, may affect the parent and child's ability to communicate (Keir, 2007; Norberg, 2010). For patients with siblings, parents reported mixed feelings about how to divide their attention between the sick child and siblings with legitimately envious feelings of being ignored (Norberg & Green, 2007; Norberg & Steneby, 2009). Daily stressors, such as preparing special meals, could also be emotionally difficult as this could produce further imbalance between siblings (Norberg & Steneby, 2009).

Supporting the child's peer relations was also indicated as an important factor for parents, and helping the child explain their diagnosis and treatment to friends, as well as helping them cope when considering future hospital appointments or physical indicators of cancer, for example, baldness, could be difficult to manage. Some parents believed that their own social network shrunk, so that some friends disappeared whilst others became closer. Feeling overwhelmed with changes in the support system led to these parents feeling emotionally overwhelmed (Norberg, 2010).

Parents linked psychological and physical exhaustion with susceptibility to stress and sleep disturbances (Wideheim et al., 2002; Norberg & Steneby, 2009). This was reported to result in taking time off work as well as feeling low in mood. Depressive symptoms were pervasive across studies, with approximately one quarter of participants scoring within the clinical range for depression (Ownsworth et al., 2009). However, since the majority were not illustrating depressive symptoms, this may conversely argue for relatively successful adjustment to the caregiver role.

Burnout is problematic because it is not only detrimental to the parent's mental health but can subsequently affect the level of care provided to the child, and reduces the capacity for effective parenting, as well as the ability to take in medical information and adjust to the role of caregiver (Norberg, 2010).

Residual stress from diagnosis and treatment

There was accord between many of the studies that unresolved stress at time of diagnosis and treatment does not gradually dissipate for many parents (Wideheim et al., 2002; Freeman, O'Dell, & Meola, 2004; Bonner et al., 2007). Instead, anxiety and trauma-related stress reactions can be pervasive, affecting the parent's coping and ability to be a caregiver for a long time after treatment.

Emotional-coping has been shown to be detrimental compared to other forms of coping, such as problem-solving, as it can lead to maladaptive cognitions that the parent is somehow to blame or that they have not coped well enough, or are avoidant and distant (Fuemeller, Mullins, & Marz, 2001; Wideheim et al., 2002; Hutchinson et al., 2009; Bennett et al., 2013). Intrusive ideational thinking about the inability to cope only perpetuates this vicious cycle by creating more pressure and stress on the carer, rendering their ability to care for their child even more limited (Bruce et al., 2010).

A separate feature, having an external locus of control has also been shown to lead to higher stress due to appraisals in forces removed from the parent-child relationship rather than an internal locus, which would focus more on the capabilities and capacity for effective parenting (Bennett et al., 2013).

There is also a widely held view within these studies that witnessing a child be treated for a brain tumour is akin to a traumatic experience and, thus, many studies included measures to identify post-traumatic stress symptoms, such as the Impact of Event Scale (Bonner et al., 2007; Hutchinson et al., 2009; Bruce et al., 2010). All of the studies investigating post-traumatic stress found that a large proportion of parents met clinical significance, for example, 43% met DSM-IV criteria for post-traumatic stress disorder in one study (Fuemeller et al., 2001). However, Norberg and Green (2007) question whether the on-going and changing stressors mean that this stress can truly be classified as *post-traumatic*.

The overarching theme for residual stress reactions has been hypothesised by some researchers as stemming from grief and/or unresolved sorrow, leading to anxiety, overcompensation of emotional-coping and post-traumatic stress (Wideheim et al., 2002; Bonner et al., 2007). This may in turn come from unmet informational needs, as well as unprocessed experiences during diagnosis and treatment (Freeman, O'Dell, & Meola, 2004; Norberg & Steneby, 2009; Norberg, 2010).

Future-oriented uncertainty

Whilst the review by Vrijmoet-Wiersma and colleagues (2008) also focused on uncertainty as a predominant factor of stress, it considered the uncertainty of relapse alone, whereas ten of the fourteen reviewed studies considered future-oriented uncertainty as relating to two main features. As mentioned, future-oriented uncertainty, the ambiguity and unpredictable nature of relapse and recurrence, was a focus of many parents' fears about their child's wellbeing. The feature did not change depending on

time points, suggesting that, whether during treatment or in the survival period, worrying about the future remained constant (Norberg & Green, 2007; Bruce et al., 2010). Furthermore, one of the biggest uncertainties regarded tumour recurrence and the inability to control or predict this outcome (Wideheim et al., 2002; Norberg, 2010; Shortman et al., 2012). This finding links with the earlier section relating to emotional exhaustion and locus of control (Wideheim et al., 2002).

A second aspect of uncertainty related to potential changes in the children's neurocognitive, emotional and behavioural functioning (Fuemeller, Mullins, & Marz, 2001; Freeman, O'Dell, & Meola, 2004; Hutchinson et al., 2009; Bennett et al., 2013). How the children may continue to change in the future, as well as a parent's inability to know what they should expect for emotional, social, physical, academic and occupational outcomes were prominent sources of stress (Wideheim et al., 2002; Norberg & Green, 2007; Norberg, 2010).

Refutational factors

Due to small samples and opposing views across studies it was difficult to form judgements about some factors that may influence and perpetuate parental stress. Whilst one of the studies found that marriage was predictive of lower distress, another study found the opposite; marriage might have protective factors, such as being able to share the burden of physical and emotional caring tasks as well as alleviating financial pressures, yet differing parenting styles may lead to conflict (Freeman, O'Dell, & Meola, 2004; Bonner et al., 2007). The majority of research suggests that as fathers take on caregiver roles as well as employment and parenting roles, they will take on stress in the

same way as mothers and will benefit from the same level of care and consideration in psychosocial support (Fuemeller, Mullins, & Marz, 2001; Bonner et al., 2007). However, one paper argues that mothers suffer higher levels of stress than fathers irrespective of the heavier parenting burden expected of mothers (Norberg, 2007).

Most of the studies did not find differences in stress levels due to time since diagnosis and treatment and that, as previously mentioned, stresses due to future-oriented uncertainty, burden, and unprocessed and unresolved anxieties did not diminish over time (Fuemeller, Mullins, & Marz, 2001; Norberg, 2007; Norberg, 2010; Bennett et al., 2013). Definitions of the post-treatment phase of illness differ drastically between studies so it is difficult to compare studies to one another. Only two investigated the presence of different stress reactions at different phases of illness, such as unmet informational needs and feelings of helplessness at treatment compared to adjustment and fears for the future post-treatment (Freeman, O'Dell, & Meola, 2004; Hutchinson et al., 2009). However, the studies' small sample sizes, broken down into further smaller groups for phase of illness-related methodologies, make outcomes difficult to validate. In addition, both were between-study designs and compared different participants, demonstrating a shortage of available longitudinal data.

Summary

The findings of this review indicate that the effects of a childhood brain tumour have far-reaching implications, not only for the patient and their on-going health status but also for the caregiver. Kazak (2005) promotes the idea of paediatric neuro-oncology as a family disease and that attention should be given to all members of the patient's system.

DISCUSSION

The dynamic interpersonal nature of the parent-child relationship means that stress reactions, such as burnout, will not just affect a parent's wellbeing but their capacity for parenting, which underlines the importance of this research.

Despite the scarcity of literature, there is enough evidence to present the case that, for many parents of children treated for a brain tumour, stress is a common occurrence that can manifest itself in a variety of ways with considerable overlap over the given domains. One clear example is that physical and emotional exhaustion were closely related to adjustment to the caregiver role. Other links have been made in the evidence base, for example, emotion-focused coping has been shown in other populations to prolong post-traumatic stress symptoms, and increase general levels of distress (Miller et al., 1996; Bryant et al., 2000).

Stress reactions from childhood brain tumours are comparable to other cancers, notably for post-traumatic stress and anxiety. Whilst the review by Vrijmoet-Wiersma et al. (2008) found relapse uncertainty a major stressor, brain tumour literature specifically highlighted future-oriented anxiety about potential neurocognitive and psychosocial deterioration (Carpentieri, Waber, & Pomeroy, 2003).

Limitations of the Review

There were several limitations in this review. Confounding variables, such as tumour type, child and parent age at diagnosis, and treatment effects were problematic to explore due to limited available data or small sample sizes. Furthermore, the wide

assortment of quantitative measures used across the studies made for difficult analytical comparisons and aggregates.

A drawback of convenience samples, both in the quantitative and qualitative designs, is that those wishing to participate often have a personal desire to understand more or have their voice heard. This may overemphasise the level of distress within the parent/caregiver population, as they may be the people who wish to understand their distress further. On the other hand, some potential participants were excluded in a few studies due to their level of distress being too high. There may also be overestimation of the White Caucasian and high employment populations, as these were the majority of parents agreeing to participate. Financial stress in lower socioeconomic status families may amplify stress levels in these populations.

Another limitation is the dearth of researchers investigating this areas, with four of the studies included written by the same academic in Sweden. This can open the possibility of researcher and sample bias.

Clinical Utility

The breadth of measures used across the studies makes comparisons of the results more difficult. Coupled with the finding that stress is prevalent in this population and can continue long after treatment, there is a clear case for standardised assessment of the factors relating to stress, such as anxiety, strain and burden, at a number of time-points. Medical staff carrying out formal assessment with parents at time of treatment may be able to predict potential psychosocial stressors and adjustment, as well as allowing the opportunity to discuss any unmet informational needs (Hendricks-Ferguson, 2000). This

is imperative given that parents face different stresses at different time points, and the content of this support will need to be adapted to suit parents' demands (Freeman, O'Dell, & Meola, 2004). Further, it is expected that if parents can predict or understand their child's neurocognitive and psychosocial changes, they will be better placed to adjust and meet the child's needs.

Norberg (2007) argues for the follow-up of families in order to monitor any psychosocial difficulties, and it should be expected that parents would require support to deal with chronic stress, as a result of diagnosis and treatment as well as with adapting to role of caregiver both in the present and future. There is evidence that, in order to reduce burnout when offering support, parents will benefit from a combination of psychological and medical interventions (Quin, 2004).

Areas for Further Research

The review finds that stress is prevalent for parents of brain tumour patients. However, longitudinal information is lacking and it will be important to monitor these stress levels over time. As detected, stressors can affect the parents' ability to provide care for the child as well as for their own needs, and it would be helpful for mixed-method designs that can reveal relationships between quantitative levels of stress and how the participants subjectively rates this.

Results on gender differences remain inconclusive, but a recent qualitative systematic review looking at parental adjustment to childhood cancer indicates differences between how fathers and mothers cope (Gibbins, Steinhardt, & Beinart, 2012). Whether these

differences are as disparate when accounting for the brain tumour population alone remains to be seen, and should be investigated in the future.

Some of the studies within the current review concluded with a discussion of interventions for parents who are faced with stress, although much of the research into this developing field remains scant. However, initial results suggest that including cognitive-behavioural coping skills as well as psycho-education about post-traumatic stress has been shown to be advantageous for parents and siblings of other types of cancer survivors (Kazak, 2005). This was reported to allow for a sense of control and to address unprocessed emotions, and it should be investigated whether families of those surviving a brain tumour will also benefit.

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Part 2: Empirical Paper

Coping with a childhood brain tumour:

A qualitative analysis of parents' experiences

ABSTRACT

- Aims:* As childhood brain tumour prognosis improves, and research focuses on quality of life for the family, insight is needed into parents' experiences of coping during diagnosis, treatment and into the survival period.
- Methods:* Ten parents of children surviving medulloblastoma brain tumour were interviewed, and transcriptions underwent thematic analysis. Qualitative methods were used to enable accounts of participants' lived experiences.
- Results:* Analysis yielded twelve themes, organised into four domains: *Focusing on the here-and-now* in which parents concerned themselves with taking one day at a time rather than thinking about what may arise later; *Overcoming helplessness* reflected the desire to provide care-giving duties; *Different needs met across the system* included emotional bonding with other parents on the ward, whilst wanting family to offer respite; *Finding a new normal* featured in the survival period when parents reflected on new values for the family. Coping mechanisms were seen as a process, changing dependant on the time period.
- Conclusions:* Parents use a range of coping mechanisms, which occur at different phases of their child's illness. Being emotionally overwhelmed at initial diagnosis and treatment subsides to problem-focused coping during recovery. In the longer term, parents use appraisals to re-establish positive life values for the child and family.

INTRODUCTION

The past few decades have seen an increase in the survival rate for childhood brain tumours due to a refinement in medical interventions (Packer, 2008). Specifically, combinations of aggressive chemotherapy, radiotherapy and surgery necessitate bouts of hospitalisation, with treatment lasting up to two years post-diagnosis. These intensive medical treatments, in conjunction with the tumour location within the central nervous system, can have numerous implications for the child's physical and psychosocial development, with estimates of 82% of survivors exhibiting at least one problem across the biopsychosocial domains (Kahalley et al., 2012). Problematic outcomes include educational difficulties and changes in personality and social competence as perceived by parents, teachers and peers (Carpentieri et al., 1993), difficulties with executive function and other neurological capabilities (Vannatta et al., 1998; Carpentieri, Waber, & Pomeroy, 2003), and impact on mental and physical health (Zeltzer et al., 2009).

The increase in survival rates and psychosocial sequelae also poses a question about the quality of life both for the patient and the family long after treatment has finished (Norberg & Steneby, 2009). Furthermore, there is the real possibility that consequences will not emerge until long after treatment has concluded (Anderson & Kunin-Batson, 2009). Surviving for many years post-treatment does not necessitate that the tumour is in the past; families must not only wait for any late effects to arise but they must also live with the risk of tumour recurrence or relapse (Vrijmoet-Wiersma et al., 2008).

The impact diagnosis and treatment has on a child's development is not only stressful for the child but also for the parents and siblings (Alderfer et al., 2009; Moore &

Wagner, 2009). For example, there can be disruption of school attendance for siblings (Alderfer et al., 2010), maternal-child dysfunctional interaction has been found to be significantly higher (Radcliffe et al., 1996), and it has been hypothesised that witnessing a child undergo diagnosis for a brain tumour is akin to a traumatic event (Bruce et al., 2010).

Research on parents of brain tumour survivors tends to focus either on stress due to uncertainty about the future (Norberg & Green, 2007), or extra roles that result in caregiver burden and parental burn-out (Norberg, 2007; Schubart, Kinzie, & Farace, 2008). Additionally, parents have reported further physical, social, emotional and financial pressures. One phenomenological study listed a series of themes regarding distress for parents of brain tumour survivors, including a restricted family life and routine, worries about the child's future, and a more demanding parental role (Norberg & Steneby, 2009). However, most of the studies do not investigate specifics of how parents experience these stressors at different times post-diagnosis.

There is relatively scant longitudinal data available for how parents cope at different time-points of their child's illness, during diagnosis and treatment and, for those that survive the tumour, the proceeding years. Findings reveal that the longer the elapsed time since diagnosis and treatment, the lower the levels of distress and corresponding psychological difficulties, such as depression and sleep problems (Freeman, O'Dell, & Meola, 2000; Boman, Lindahl, & Bjork, 2003). Elsewhere, distress is reported to remain high, with one study suggesting similar levels of distress at six months and 18 months post-diagnosis (Sloper, 1998). Within these studies, all childhood cancers were eligible

for inclusion, with no consideration given to how long-term consequences that result from neuro-oncology treatment may affect parents when compared to other cancer types.

The majority of the studies considering adjustment to the child's status at various time points were quantitative by design. Only two qualitative studies have recently explored parents' experiences at different time points of treatment, with both concluding that coping for a child with cancer is a process (Yeh, 2003; Wong & Chan, 2006). These findings indicate how initial shock and denial subsides to acceptance and coping through practical support (e.g. care-giving and receiving information) and, finally, looking towards the future after re-establishing routines. Again, however, these samples were heterogeneous, with only one of nine parents caring for a child with a brain tumour in the larger study (Wong & Chan, 2006).

Adjustment and Coping

The psychological constructs of adjustment and coping with illness have long been prominent in healthcare research due to the focus of informing clinical practice in order to help people with a range of medical conditions and distressing life-events (Adler & Matthews, 1994). Psychological adjustment to illness refers to people's abilities to rebalance within new circumstances (Stanton, Collins, & Sworowski, 2001), and can be successful or not depending on adopting coping strategies, such as maintaining healthy behaviours, having a functional daily routine, holding high esteem, and life satisfaction (Taylor & Aspinall, 1996). These outcomes can be affected by variations in personality, cognitive adaptation and the perception of control, as well as how stress is managed through proactive self-regulation (Adler & Matthews, 1994; Sharpe & Curran, 2006).

In response to emerging research of the stressors affecting parents of children with cancer, Grootenhuis and Last (1997) carried out an initial systematic review of adjustment within this clinical population. Findings suggested that there are deleterious consequences for parents of children with cancer, including mental and physical health complications, and anxiety through fear of relapse in the future. Recent systematic reviews of parental adjustment to childhood cancer include quantitative reviews (Klassen et al., 2007; Vrijmoet-Wiersma et al., 2008) and mixed-methods reviews (Thibodeaux & Deatrick, 2007; Long & Marsland, 2011), with comparable findings. For example, parents of children with a brain tumour will need to extend their role as caregiver to meet demanding physical and emotional needs. Many researchers agree that the success of the parent's ability to cope will depend on how successfully they adjust to their child's status. At the same time, there can be positive adjustments, such as more importance placed on valuing life (Greenberg & Meadows, 1991; Peck, 1979), and reports of family bonds becoming stronger (Koch, 1985). A qualitative systematic review of parental adjustment to a cancer diagnosis was carried out in response to the heavy weighting of quantitative studies, and revealed further coping mechanisms: that parents want to feel in control, they value practical and emotional support, and that adjustment is an on-going process that changes depending on whether the child is in or out of treatment (Gibbins, Steinhardt, & Beinart, 2012).

There has been a rise in employing qualitative methods to understand the impact childhood cancer has on the family, with the implications that parents experience coping in various ways (Van Dongen-Melman, Zuuren, & Verhulst, 1997; Semple & McCance, 2010). Patterson, Holm and Gurney (2004) conducted focus groups of parents of cancer

survivors to determine three subsets of coping: emotional coping, for example humour, crying and denial; problem-focused coping by advocating for the child or giving up one's job; and using appraisals to cope, such as being hopeful and positively framing the experience of being wilful. However, as with other studies, all cancer types were included, with only 12% of those recruited surviving brain cancer (Patterson, Holm, & Gurney, 2004). Furthermore, average time since treatment completion was four years, with little consideration given to how coping might differ depending on phase of illness.

Whilst these reviews provide useful information for medical staff, patients and their families, the evidence-base argues towards marked differences between children with a brain tumour and other childhood cancers. For example, due to the location of the tumour within the central nervous system and the intensive treatment, the child's neurological and psychological development may be severely affected differently to other cancers (Packer, 2008). Reviews often incorporate brain tumours into all childhood cancer but the few studies investigating brain tumours alone reveal that parents face some atypical pressures, such as late effects of treatment (Anderson & Kunin-Batson, 2009) and higher rates of post-traumatic stress symptoms (Manne, DuHamel, & Redd, 2000). Due to this, there is a gap in the literature about parental experiences within the paediatric neuro-oncology population alone, which may identify different coping mechanisms, thus informing better clinical practice.

Medulloblastoma is one type of brain tumour, located in the posterior fossa of the cerebellum, and is more common in children than adults (Johnson et al., 1994). Although 70-80% of children are expected to reach five-year survival, treatment is intensive, combining surgery, chemotherapy and radiotherapy, and typically results in

deleterious psychosocial and physical outcomes, such as below average IQ and problems with motor dexterity (Johnson et al., 1994). Medulloblastoma was selected for this study as the focal tumour type, as it will allow for homogeneity within a population that has had to adjust to and cope with substantial changes to quality of life.

Aims

Although there is pre-existing literature about the longitudinal experiences of parents of children with cancer, there is no specific information about those caring for a child with a brain tumour. This retrospective study aimed to give a comprehensive account of the experiences of parental coping with a paediatric medulloblastoma brain tumour at different time periods, from diagnosis to survival. Findings revealed if the experiences of this population is congruent with other childhood cancers.

Most research in this field relies on questionnaire-based parental stress reactions, as well as including all childhood cancer types, thus not capturing a homogeneous brain tumour sample, and ignoring the subjective experiences of the participants. Therefore, the study will fill a gap in the childhood cancer literature by focusing on parents' qualitative experiences of coping with brain cancer specifically and how this may differ depending on the illness phase. Participants were also asked to provide their views regarding any coping mechanisms they employed and that they may wish to share with professionals and other families.

Qualitative methods are a useful way of gaining an in-depth understanding of the complex psychological and interpersonal processes that underlie effective therapeutic interventions (McLeod, 2001; Pistrang & Barker, 2010). They have an important role in

developing and modifying a richer theory grounded in data (Braun & Clarke, 2006). In contrast to hypothesis-testing approaches, qualitative methods can be inductive and allow for exploration of data, eliciting information that might otherwise go unnoticed or be taken for granted. At the same time, new or unexpected themes may also emerge.

METHOD

Design

The research undertaken was a qualitative retrospective study of how parents experience a paediatric medulloblastoma brain tumour at various time points: diagnosis, treatment, and post-treatment.

Participants were purposively sampled from a database in an inner-city children's hospital. Only including parents of children surviving medulloblastoma allowed for homogeneous sampling, and thus allowing a description of one neuro-oncology subgroup in depth (Patton, 2001). Those meeting eligibility were recruited and interviewed about their experiences.

Thematic analysis was selected to understand the experiences of parents, and was the preferred method of analysis due to its ability to encode and interpret patterns across a data set, thus enabling a description of subjective experiences (Pistrang & Barker 2010).

Ethical Approval

Approval was gained through the National Research Ethics Service on 20th June 2013, as well as through the University's Ethics Committee, the local hospital's Clinical and

Research Adoptions Committee, and the hospital's Research and Development (see Appendices C-D for details).

Participants

Participant Criteria

A clinical nurse in the hospital's neuro-oncology unit identified potential participants who met the following criteria: parents were required to have a child diagnosed and treated for medulloblastoma tumour between the primary school ages of five and eleven years so that there already existed an informed expectation of their psychosocial capabilities, the child was at least three years post-treatment to allow for any treatment effects to be present, and the child was alive and had no current relapse or recurrence.

Parents were also required to be able to provide informed consent, and speak English.

Procedure

Thirteen prospective participants were purposively sampled from the hospital's neuro-oncology electronic database using the inclusion and exclusion criteria. A member of the clinical team, telephoned to inform them about the purpose of the study. Following this, an information sheet was posted (see Appendix E), and interview times were booked during a second phone call by a member of the research team.

The participant information sheet was read through again at the interview and any questions were discussed. Participants were then handed a consent form before

beginning the interview (see Appendix F). Consent was discussed and, once agreed, the participants signed the consent form.

All participants were informed in the information sheet sent in the post that they would receive a £10 gift voucher for their time, funded by the University. The clinical team agreed that the process must be handled sensitively and it was decided that the vouchers would be given to participants following the interview, with the researcher explaining that they were a small token of appreciation.

Sample Demographics

Data collection commenced in August 2013 and continued for three months. Of the thirteen families meeting the inclusion criteria, ten agreed to participate. One family moved away and was unable to be contacted, another prospective participant's child had tumour recurrence during recruitment, and the third decided not to participate at the time of the initial phone call.

Table 2 shows participant demographics; to ensure anonymity, all names have been modified and any identifying features removed. All participants chose to be interviewed at home rather than at the hospital. All of the interviewees were female and ranged in age from 30 to 49 years at the time of study, with the mean age being close to 42 years of age. Six of the families were White British. Prior to diagnosis, five of the participants were employed and one worked part-time work. As a result of the tumour, all but one of the working mothers reduced their working hours or stopped working altogether.

Of the ten participants, all of their children were diagnosed with medulloblastoma and were treated with surgery, chemotherapy and radiotherapy, lasting for up to eighteen

months post-diagnosis. Children were between the ages of 5 and 11 years at time of diagnosis with a mean age of 7 years, and a mean of just under 5 years since completing treatment. The diagnosed children were aged between 9 and 17 years of age at time of recruitment, with a mean age of 14 years. All had consequences of diagnosis and treatment, with common difficulties including slow processing speed, and problems with gait and self-care.

Table 2. Participant Demographics

Participant	Age at Study	Ethnicity	Child's Age at Diagnosis	Child's Age at Study	Impact of Tumour (includes but not limited to)
P1	40s	White British	10	17	Gait, memory, processing speed
P2	30s	Asian	8	16	Intellect, processing speed, self-care
P3	40s	White British	7	16	Gait, processing speed, social skills
P4	30s	Black British	7	12	Memory, mood, social skills
P5	40s	White British	5	14	Intellect, memory, physical, self-care
P6	40s	White British	5	9	Gait, memory, processing speed
P7	40s	White British	8	13	Hearing, mood, processing speed
P8	40s	White British	5	15	Memory, self-care, visual
P9	30s	British Asian	7	12	Gait, intellect
P10	40s	White European	11	17	Memory, processing speed, self-care

Interviews

A semi-structured interview schedule was constructed following discussions with the clinical team and their previous experiences working with parents who had experienced a child with a medulloblastoma. It was agreed that the framework should focus on parents' experiences of coping in relation to changes to the child's physical and psychosocial functioning. Research by Patterson, Holm and Gurney (2004) indicates that parents utilise a multitude of coping styles, and the clinical team decided that the interviews should be organised into three distinct time periods to determine whether coping styles vary by time or occur with no clear pattern: experiences pre-diagnosis, experiences during diagnosis and treatment, experiences following treatment, as well as any general thoughts about coping (see Appendix G for full schedule). Flexibility in structure was used with the aim of allowing participants to talk from their own frame of reference.

Due to the sensitive nature of the conversation, time was given at the beginning of the interview to engage the participant and explain the rationale. Following this, the consent form was signed. Clear guidance was given that participants could opt out at any time without affecting any future standard of care and that, if they were distressed by the discussion, a member of the hospital's paediatric psychology team would be available to meet with them.

Time was also set aside at the end of the interview to ask whether the participant wanted to append or reframe any of the discussion points. Participants were thanked for their time and informed that the interviews would be transcribed, kept confidential and made anonymous. As one of several credibility checks within this study, taken from an

established framework for conducting qualitative methods (Spencer et al., 2003), participants later had the opportunity to read over their own transcript to check for identifying information as well as correcting anything they felt was misinformed.

Analysis

Coding

Interviews ran from fifty to eighty-five minutes in length and were recorded on data protected equipment. No one opted out of the study. All participants were offered to clarify or add anything at the end of the interview, as well as asked to reflect on the experience of the interview.

Interviews were subsequently transcribed and read for total immersion of the data. A second researcher, a trainee clinical psychologist with experience in qualitative analysis, read eight of the ten transcripts to allow for a consensus approach. Throughout the coding process, the researchers maintained communication to clarify themes.

The thematic analysis involved noting any preliminary themes or ideas in the margins of the text (Braun & Clarke, 2006). A second reading fine-tuned these preliminary ideas into a thematic framework, which was then systematically applied to the other transcripts in order to identify patterns in the data (see Appendix H for an example of an analysed transcript). The second researcher also noted whether these themes were present. Following this, both researchers met and had to agree that themes appeared in transcripts in order to include them.

The analysis was sequential, with the data appearing to hit saturation before the final transcription. Recurring themes were labelled as subordinate themes and clustered into coherent superordinate domains. Themes were then checked to ensure that the original data was vivid and meaningful. Figure 2 shows a diagrammatic representation of the data analysis.

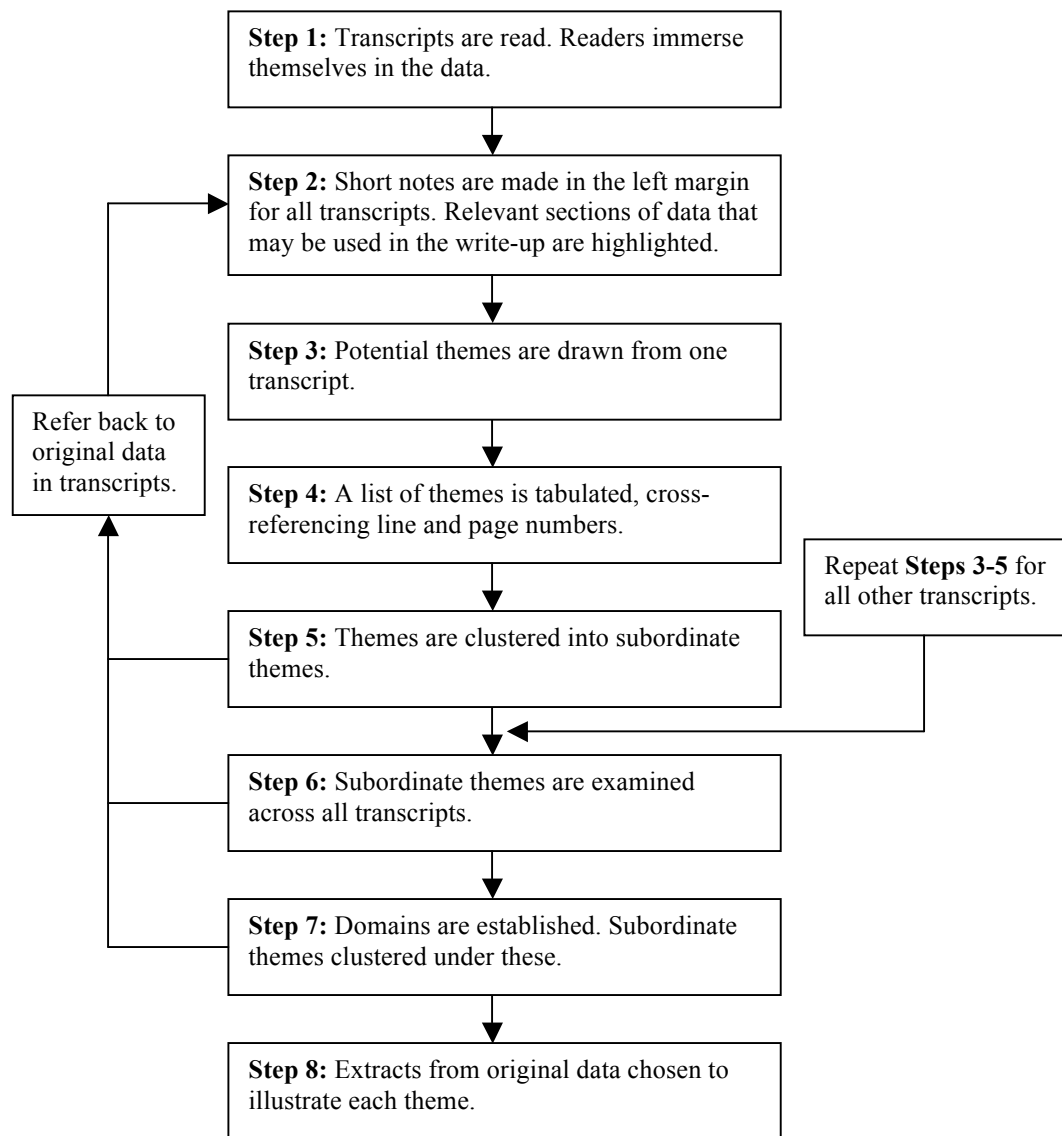


Figure 2. Diagrammatic representation of the data analysis

RESULTS

Organisation of the themes

The analysis generated twelve central themes, grouped into four domains (see Table 3); Appendix I reveals a breakdown of subthemes within a mind map. There is an emphasis on what is happening day by day in the earlier stages of diagnosis and treatment, when survival is still uncertain, leaving adjustment to long-term psychosocial outcomes at the back of the mind (*Focusing on the here-and-now*). Once a treatment routine sets in, parents turn to offering help (*Overcoming helplessness*), and look to social networks for support (*Different needs met across the system*). Only later, when a child's survival is more promising and developmental changes are more pronounced, are parents able to reflect on what has occurred (*Finding a new normal*).

Although physical and psychosocial outcomes varied in severity, participants generally held similar views about their experiences and how they coped at different time points. This is especially true of the initial shock at diagnosis, having to put their own needs on hold to cater for their child, and that all the participants, regardless of how they perceived their child's abilities, were absolute in that they now wished for their child to be happy above all else. Table 4 reveals the frequency of themes emerging in the data.

The following is an exploration of these themes, organised by domains. Direct quotes from participants are used to support the findings by grounding them in participants' accounts and provide resonance with readers' understandings (Spencer et al., 2003).

Table 3. Domains and themes

Domains	Theme
Focusing on the here-and-now	The start is a whirlwind Take one day at a time Every child is different
Overcoming helplessness	Concentrating on physical recovery Give up everything else Keeping in touch with the school
Different needs met across the system	Staff to be available but not overbearing Talk to others in the same boat Accept practical help from family and friends
Finding a new normal	Grieving for lost opportunities Following the child's lead Hope for the future

Focusing on the here-and-now

The symptoms before a brain tumour diagnosis can be stressful for parents, who are often upset by the misdiagnoses before the tumour is revealed. Whilst diagnosis can take months, treatment is immediate, and leaves little time to process what is happening other than to concentrate on the immediate health of the child, day by day: “You’re on a treadmill. I don’t think we ever had a massive breakdown, I think you get swept up in this...it’s all very surreal, you have to just keep going.” (P6). Consequently, parents only want to think about their child’s survival and are unable to take in information about long-term treatment plans or lasting effects: “You survive today. Don’t worry about tomorrow. You survive tomorrow. Don’t worry about the next day” (P8).

Table 4. Frequency of themes by participant

Theme	P1	P2	P3	P4	P5	P6	P7	P8	P9	P10	<i>N</i>
The start is a whirlwind	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	10
Take one day at a time		✓	✓	✓	✓	✓	✓	✓			7
Concentrating on physical recovery	✓		✓	✓	✓	✓	✓				6
Give up everything else	✓	✓	✓	✓	✓	✓	✓		✓	✓	9
Keeping in touch with the school	✓		✓	✓	✓	✓				✓	6
Staff to be available but not overbearing	✓	✓		✓	✓	✓	✓	✓		✓	8
Talk to others in the same boat	✓	✓	✓	✓		✓		✓		✓	7
Accept practical help from family and friends		✓	✓	✓	✓		✓	✓			6
Grieving for lost opportunities	✓				✓		✓	✓	✓	✓	6
Following the child's lead		✓	✓		✓	✓			✓		5
Hope for the future	✓	✓	✓	✓	✓	✓	✓		✓	✓	9

The start is a whirlwind and parents must sit by as medical teams go to work. Several of the participants retained statistics concerning survival rates (three year survival at approximately 83%) but they took in little else other than a day-by-day account of their child's health. Four parents made explicit use of the word 'whirlwind', with phrases such as "the whirlwind I was in" (P1) to describe initial treatment.

During early stages of treatment, parents acknowledge that decline and tumour recurrence are possibilities but are hesitant to think too much about the future. A shared coping mechanism was to **take one day at a time**: "With brain tumours there are so many bad outcomes that can happen, and not all of them likely to happen to every child, and to be told all of those bad things on day one when you're trying to deal with surgery it would be too much and some parents would collapse under the strain" (P3). All of the participants shared the experience of wanting to focus solely on that day's treatment, seeing little purpose in ruminating about potential developmental changes in the future when the immediate days were precarious and the outcomes uncertain. Only in latter stages of chemotherapy were most participants ready to consider long-term consequences: "You need to worry far enough ahead to put things in place but don't worry unless you need to" (P8). During this time, it appears beneficial to cope by being emotionally guarded from thinking about the future: "I just thought about getting through it. This bit is hard enough. I didn't think long-term. I wasn't thinking too far ahead. It's probably best not to. You concern yourself with what is going on in the here-and-now. I just thought at least she is up and alive" (P7).

A common mantra held was that **every child is different**. Participants saw little purpose in comparing their children to others on the ward when outcomes are varied and

survival or death is uncertain: “My son is getting this treatment the same as other children, but he’s reacting differently so what I want for him they might not want for their child” (P4). Talking to other parents on the ward satisfied the participants that children are individuals who share the same diagnosis and treatment plan but potentially little else.

Overcoming helplessness

Parenting roles can be diminished when intensive medical intervention is needed and parents reported feeling helpless, considering much of their time was spent in and around the hospital whilst waiting on updates. Once the child was more stable, parents could be hands-on with care-giving. This typically meant that all of their efforts were spent caring for a sick child but often to the detriment of their own emotional and physical needs: “Every time he vomited, his tube had to be reinserted and was a nightmare, and ended up with a regime where the pump had to be adjusted several times a night. So every night I was woken repeatedly and I wasn’t getting any sleep during the day. I couldn’t cope” (P3).

Although asked about psychological, social and educational trajectories, six of the participants reported that, first and foremost, **concentrating on physical recovery** served as a coping strategy by means of seeing tangible improvements in the child’s quality of life, but also by distracting from other concerns: “There wasn’t a lot I could do but get him back on his feet. We did the physiotherapy, did the exercises, then we’d worry what will come later” (P1). Physical recovery was also a demonstrable measure of improvement: “Initially, we concentrated on the physical side of things. It was

impossible to tell what had happened intellectually as he couldn't speak... and it took a long time for the academic issues to be revealed... we only realised he had stepped back a long way when he returned to school" (P6).

Of the six employed participants, five cut back their hours. The caregiver role becomes all-consuming as parents **give up everything else**: "I had to stay at home all the time and not go out" (P2). Parents not only sacrificed their own health but their careers, hobbies, and relationships with other family members, most notably any of the patient's siblings. All but one participant reported that coping becomes less about emotional well-being and more about doing everything to keep the child well: "Initially, it was shutting things away. I think because he got up, we got up. I suppose there were lots of mornings we didn't want to get up but we had to. Caring for him was a full time job, co-ordinating everything. Your whole life is occupied" (P5).

Participants recommended that parents should maintain an aspect of life that is separate from their child: "I'd want [parents] to take time for themselves because... we focus on getting the child better and forget about ourselves and it gets to you as well. You neglect yourself, which is not good because it affects the child" (P4). However, participants admitted that, when the child requires constant care, it was something they struggled to follow through themselves: "I think one other thing I'd recommend but I didn't manage to do it is to keep your life going, whatever it is that keeps you as a person... I managed to keep my voluntary work going and it was a lifesaver. Having that position where I'm not 'his mum' has meant an awful lot to me. Keep one thread and prioritise it" (P3).

Keeping in touch with the school was another method for parents to feel helpful.

The less time and distance the child felt from the school the more likely participants believed he or she would assimilate on return: “The reception teacher used to come up every single week to see him in the hospital, and she would bring paper chains or the like, and kept that link with school” (P6). The various suggestions for this included: updating teachers, adapting the school setting to meet the child’s physical and intellectual needs, integrating the child socially, and normalising the physical appearance to classmates.

Parents also found value in asking hospital staff to provide education to the schools: “I think fear is a big factor. One nurse practitioner talked to the kids in the class and that was excellent because he was in the wheelchair and on a tap, with no hair and looked quite terrifying, and it was really beneficial as they then knew what to expect” (P3).

Different needs met across the system

Not all support is welcomed equally by parents: “Can’t say I spoke much with those who didn’t understand... those who weren’t in it. In the hospital if I met someone actually going through it, that’s more support from there. It’s a very private, very unique thing” (P8). Three different groups, being able to offer different types of support, emerged from the analysis: staff, other parents on the ward, and family and friends.

All participants valued the medical expertise of the hospital staff but the focus was on **staff being available but not overbearing** with information: “They might have said she won’t be able to walk properly in the future but I would not have taken it in until later on. I don’t know if, had they told me everything, I could’ve coped, or whether it would’ve made it worse” (P2). Although participants didn’t want information held back,

especially when presented alongside decisions about care, anything other than discussions about short-term treatment was not always welcomed. Participants wanted updates in small doses; being pointed towards research in leaflets and online allowed them to absorb the information at their own pace.

Participants were grateful when staff knew them by first name and were assigned a care-coordinator, welcoming consistency and being displeased at having to explain the back-story to new workers: “We see physiotherapists and know most of them and they know when there’s been a change. What bothers me now are the changes in the NHS and cutbacks. Then you get new staff and it’s making it harder because I have to sit there and talk about the past again” (P5).

Participants also learnt to seek help from the wider community. Participants were advised to ask staff about organising financial support, such as state benefits, with haste; the earlier stages of adjusting to the child’s needs were reported to be the most difficult and, unless informed help was organised quickly, it often came too late.

Private rooms at the hospital were appreciated but there is a trade-off in loneliness. The flipside of this, being on the ward, meant a lack of privacy but came with reciprocal support with other parents; at its most basic, “they told us what happened with their children and we told them” (P2). The majority of participants preferred to **talk to others in the same boat**, and had their emotional needs met from those who shared their experiences: “You share with people who understand. Even if it’s different, just the idea of a kid going through the same treatment, you feel someone’s there to understand you” (P8).

Interviewees did not merely offload their issues but consciously supported one

another, resulting in life-long friendships: “We get together for lunch and it’s been positive talking about the past. We did things like bought a bottle of Pimm’s between us when one was going home after chemo. It seemed decadent but why not when you’re stuck in hospital” (P3).

Over half of the participants relied on family members and friends to help with the logistics of being at hospital instead of at home with the other children. However, many felt uncomfortable talking within this network, deeming the stigma of ‘brain tumour’ and treatments such as ‘chemotherapy’ as alienating and distressing. At first, parents found it difficult to **accept practical help from family and friends**, believing they should be coping on their own. Instead, of turning to them for emotional support, which they received on the ward, parents learnt to ask for respite: “When you first begin you think you can cope and you can’t and sometimes you miss out on opportunities. Things like providing support to siblings is quite an easy role for friends and family to do. Take them out and give them a life, which you can’t do because you’re too busy worrying about the other child” (P5).

Finding a new normal

The interviewees reported that they came to understand that a brain tumour may have an acute onset but the repercussions are chronic. Treatment is lengthy, check-ups occur far into the survival period with the fear of tumour recurrence, and developmental changes can affect the child and the family for the rest of their lives: “He’s had this thing happen to him and now he’s got all this as well. We’ve applied for his driving licence but I need to get his medical records, so we’re looking at another twelve weeks

before they'll assess, so it's still affecting him. It will be never-ending" (P1).

Adjusting to a child's new capabilities is a lengthy process, as detecting changes takes time and, earlier on, parents have neither the energy nor desire to think long-term. Although parents acknowledge detrimental changes, they can also make positive appraisals: "There's no reason he shouldn't have a perfectly good career. Whilst he may never work full time, he doesn't allow anything to get in his way and I think he will find someone to employ him and get a lot out of him" (P3).

Some participants spoke with negativity, **grieving for lost opportunities** that their children had missed out on, such as hobbies with peers and career prospects: "I just can't think what she possibly could do. She's going to need to have something to be involved in but I can't see how she can do it on her own. I don't mind being at home... but it does bother me I've had that choice taken away, and having choices taken away for her as well is hard" (P7).

However, parents also spoke with optimism; four of the participants stated that **following the child's lead** was a constructive way of adjusting by exchanging disappointment for pride: "His positivity influenced me. When I saw him do positive stuff, like drawing, I thought it's not the end of the tunnel so why am I sitting here getting emotionally sick whilst he is still smiling?" (P9).

When survival was deemed more likely, parents were able to restore their optimism and had **hope for the future**, reappraising expectations of their children's prospects. Although some parents held a lingering sadness that the child would not achieve what they could have had the illness not occurred, most saw improvements in health as a top priority: "They've gone through a big thing in life and when they come out it's a good

outcome. You see them walking and talking and as long as they can communicate with the world that's perfect, you don't need anything bigger than that" (P10).

All of the participants reappraised the value they placed on life. For many, the illness had been an opportune time to adjust things they weren't happy with, such as spending more time together. Regardless of past expectations, parents shared the same intrinsic hopes for their children: "We want him to be happy, content and included. We've been looking around secondary schools and met the SENCO. Her last words to us were, 'Don't ever have any limits on your aspirations for him.' I don't think we'll ever forget that" (P6).

Experiences as a process

Although participants did not explicitly spell out a timeframe for a clear distinction between when diagnosis ends and treatment begins, or how far post-treatment they considered the survival period to begin, it appeared that themes were more relevant to participants at different time points, and that their experiences of coping varied accordingly. For example, there is more emotional coping earlier on, with denial and shock at diagnosis, whilst problem-solving coping becomes more meaningful when the child is recovering, and then only later are appraisals made.

Not all of the themes are easily aligned within different time periods and there may be overlap. Figure 3 shows an estimated timeline of when the themes may present themselves according to when they typically emerged in the interviews.

	< Pre-diagnosis	>× Diagnosis <×	Treatment	>×	Post-treatment and Survival
Focusing on the here-and-now			The start is a whirlwind		
			Every child is different		
			Take one day at a time		
Overcoming helplessness			Concentrating on physical recovery		
			Give up everything else		
			Keeping in touch with the school		
Different needs met across the system			Staff to be available but not overbearing		
			Talk to others in the same boat...		
			Accept practical help from family and friends		
Finding a new normal					Grieving for lost opportunities...
					Following the child's lead...
					Hope for the future...

Figure 3. Themes when they occur on the timeline, organised by domains

DISCUSSION

Caring for a child who is treated for a brain tumour, and dealing with the physical and psychosocial after-effects, can be distressing for parents; not just during early diagnosis and treatment but at much later phases. The findings replicate other studies that suggest that parents suffer a range of stress reactions and use various coping mechanisms (Yeh, 2003; Wong & Chan, 2006). The present study raises the important implication that parents alter their coping styles by phase of illness. Parents are more likely to emotionally shut-down, “making it through the storm” (P5), at the initial stages of diagnosis and treatment when they feel helpless, whilst adopting more practical roles when the child is recovering from the more rigorous treatment regimes. From then, parents are able to plan for consistency in routines and help enable the child to regain normality in everyday activities, such as through links with schools and peers. Only later do parents use appraisal-based coping to re-evaluate their hopes in life.

One of the aims of the study was to focus on experiences of parents of children with a brain tumour separately to other cancer types, as previous research tended to view the cancer type as indistinct (Van Dongen-Melman, Zuuren, & Verhulst, 1997; Semple & McCance, 2010). Many of the findings derived from this population are congruent with other studies investigating paediatric brain tumours as well as other childhood cancers. For example, burden, burnout and other stress reactions appeared to be present for many of the participants, although noted observationally and not measured through standardised questionnaires (Norberg, 2007; Vrijmoet-Wiersma et al., 2008).

Participants were explicit that they didn't want to be overwhelmed with medical information, instead wanting to learn the specifics of the diagnosis and treatment plan over time and through extraneous sources outside of medical meetings, such as reading leaflets and websites. Although parents knew the severity of the diagnosis, few described how this compared to other illnesses. This is especially true for the specifics of medulloblastoma, with only one parent mentioning the differences between tumour types in the interviews. This may be due to the speed at which treatment begins, in which parents seldom have the time or emotional headspace to learn about this. Furthermore, parents see their own child as unique so, although some consequences to quality of life are known, there is little forward-thinking until treatment effects are present; by this point the specifics of the illness may not be of prime importance.

Existing literature suggests that parents use many of the same mechanisms of coping as with other cancer types, but these were not given in relation to phase of illness (Patterson, Holm, & Gurney, 2004). The findings of this research suggest a process of coping and adjustment: at first, coping is largely passive, when parents embrace shock and denial and "getting through it" (P7). After some adjustment to the illness status, this subsides to problem-focused coping mechanisms, such as providing care-giving, as well as a shift in emotional coping from denial to sharing stories with other parents on the ward. Finally, appraisal-based coping begins to happen when there is optimism about survival and re-evaluation can take place. Examples of the strategies parents reported using at different phases of illness can be seen in Table 5, using Patterson, Holm and Gurney's model of coping (2004).

Table 5. Strategies employed, by coping types (Patterson, Holm, & Gurney, 2004)

Coping Type	Strategy Employed by Parents	Expected Phase of Illness
Emotional	Shock, denial	Diagnosis, Early Treatment
	Sharing experiences with ward parents	Treatment
Problem-Solving	Physical care-giving	Treatment
	Keeping links with the school	Treatment
	Filling out applications for support	Treatment
Appraisal-Based	Following the child's lead	Treatment, Post-treatment
	Establishing new life values	Post-treatment

Although the sample were all parents whose children had survived, the findings are relevant to all parents of childhood brain tumours during initial diagnosis and treatment, due to a shared uncertainty of survival at this phase. Participants did not want to consider long-term consequences and, to begin with, staff may be better off concerning themselves with offering practical support and be mindful that parents may not be emotionally available to think long-term. Only later, during recovery and survival would conversations about practical support and appraisals be implemented.

Staff members were praised by parents during the interviews for their genuine support, especially those who were consistently present at hospital visits and knew the family well. Further appreciated facets of staff roles included liaison work with schools and helping to set up other types of support for when the child became an outpatient; whether filling in forms for disability allowances or troubleshooting practical setups around the house. This practical support has been documented as a factor towards

parental coping during child illness elsewhere (Hopia et al., 2005; Sarajarvi, Haapamaki, & Paavilainen, 2006).

Whilst problem-focused coping occurs, other parents on the ward can offer emotional support by sharing their own advice, having “been in the same boat” (P8) and highlights the benefit of informal social support (Compas et al., 2012; Coulson & Greenwood, 2012).

Participants were divided as to whether the well-reviewed resources at this particular hospital, such as private rooms, meant that something about kinship with other parents was lost. The hospital hosts coffee mornings, but parents asked if staff could facilitate more opportunities for informal time with other parents. This is not always feasible, considering parents did not wish to leave their child’s side, but were appreciated when nursing teams offered this respite: “Sometimes you need to leave the bedside and the nurses are there to step in” (P3).

Feeling helpful was something that participants wanted to achieve but acknowledged that it can also be troublesome, as there is ambivalence between knowing that one’s own physical and emotional needs can be lost when caring for a sick child. Participants were clear that they wanted to hold on to some personal aspect of their life, outside of the care-giving role. It may be valuable for staff to support parents by gently encouraging them to consider this.

The study benefitted from a qualitative method, as it allowed participants to describe how they experienced caring for a child in a way that wouldn’t be captured through standardised measures that tend to focus on stress reactions (Freeman, O’Dell, & Meola, 2004; Bennett et al., 2013). Furthermore, the analysis did not concentrate solely on the,

more-negative, stress reactions that typically feature for research on this population, but also opened up discussion for positive appraisals; congruent with evidence that post-traumatic growth allows for positive change following trauma and adversity (O’Leary & Ickovics, 1995; Hefferon, Greal, & Mutrie, 2009). It was encouraging to learn that even in the face of anxiety about future-uncertainty, grieving for lost opportunities during childhood and worrying about future employment and relationship possibilities, parents were able to shift their expectations and re-establish new hopes and aspirations: “He doesn’t see himself as being disabled. He doesn’t remember anything about his past before his illness. Since then everything has been an improvement and, because he has continued to make progress, he feels like everything is going well. He doesn’t see any disappointment about what he’s achieved... I have readjusted” (P3).

Positive values were ascribed to what parents hoped for their child but also for the family as a whole. For example, one participant’s husband cut back his work hours to spend more time at home, whilst another participant booked a holiday that she had been delaying for many years, deciding to “do what I’d put off year after year” (P4). This ability to attribute new, optimistic values and life satisfaction is congruent with the experiences of parents in a qualitative review of all childhood cancers (Gibbins, Steinhardt, & Beinart, 2012).

The finding that parents use a multitude of coping strategies is consistent within the literature (Kazak et al., 2005). However, this study progresses the knowledge base by revealing that coping differs depending on the phase of the child’s illness, synonymous with other research on coping as a process (Amato, 2000; LaMontagne, 2000; Padden, Connors, & Agazio, 2011). By mapping the experiences parents have, coping can be

viewed as an evolving process, from “battening down the hatches” (P5), to re-evaluating what is wanted from life: “I just want my child to be happy” (P6).

Limitations and directions for future research

Although the study was experiential and it can be hypothesised that it is relevant to many other families, caution should be exercised when generalising the findings beyond the population. Due to all participants having good experiences with the hospital and its resources, and including only families of children who survived, a positive bias cannot be ruled out. It is hoped that the flexible interview structure and impartiality of the interviewer would allow participants to be open about their experiences, and it appears that the majority were able to positively reframe their lives despite changes to their child’s quality of life.

The aim of the study was to determine parents’ experiences at various time points following diagnosis and, therefore, interviews provided retrospective data. Although necessary for the design, retrospective interviews require the interviewee to remember distressing times and being able to express complex internal processes (Giorgi & Giorgi, 2003). Furthermore, many participants acknowledged that only years later did they feel comfortable reflecting on the past: “If you’d have asked me a couple of years ago to take part I would have said no. I wasn’t ready to look back” (P6). Whether participants’ memories, therefore, are an accurate portrayal of what happened during earlier phases is something that should be viewed with caution when using the findings to inform clinical practice. Interviews with parents at various time points to compare their experiences and

determine whether coping strategies differ accordingly could circumvent retrospective drawbacks, although recruitment during these stressful times may be more problematic.

As a purposive sample, ten families were recruited with all participants being mothers. Recent research shows gender differences in parents experiencing a child with a brain tumour (Gibbins, Steinhardt, & Beinart, 2012), although stress levels between genders have also been reported as comparable (Bonner et al., 2007). Nonetheless, differences between mothers and fathers in terms of emotional and physical burnout suggest that future research studying parental responses to having a child with a brain tumour may benefit from including fathers. Within families with two parents it is often the case that one parent, typically but not always the mother, acts as primary caregiver whilst the other performs the roles of financial supporter (Bonner et al., 2007; Norberg, 2007). Perhaps comparing the experiences of parents by establishing the proximity to care-giving duties may reveal intricacies in how coping styles may vary further.

The present study points towards congruence with other research that explores parental experiences of childhood cancer, and sheds light on adjustment being an on-going process. Whether this process is true for all cancer types and, for that matter, all childhood illnesses, is something to research further. Future studies should compare these populations, enabling better understanding of how parents experience coping with a child during illness and recovery. As most of the research in this field is quantitative, this study has illustrated the value of incorporating an inductive approach to complement outcome-based designs, drawing from the parent's frame of reference directly, and reveals information about the complexity of experiencing childhood cancer that would be sparser when employing quantitative methods alone.

Clinical Implications

Pertinent from this research is the importance for staff to consider parents' experiences of coping as a process, as seen in Figure 3. Following treatment, parents may be able to integrate any changes in their child's physical and psychosocial functioning through practical adjustments to the daily routine as well as making appraisals about their experiences. Patterson's Family Adaptation and Adjustment Response Model (1988) indicates that at this point, parents may not be under as much stress as during earlier treatment and thus have the capacity for managing any strains through applying coping tools, such as accepting social support and problem-solving logistics of childcare.

However, during initial diagnosis and early treatment, emotional strains, such as a focus on their child's illness status, may overwhelm the ability to problem-solve or make appraisals. This finding fits Patterson's model (1988) and is also in line with Lazarus and Folkman's Transactional Model of Stress and Coping (1984) since stressors, such as the child's illness and the parent's feeling of helplessness, may outweigh the parent's ability to manage. For example, as participants explained, their own health may be at the detriment of caring for the child.

Parents may only think in the short-term at early stages of treatment, and staff may discover that attempting to offer information other than immediate updates about the child's current health is not retained. Participants in this study suggested that, although they are grateful for the availability of future-oriented information, they would prefer to choose when to receive it. Whilst some decisions, such as treatment plans will need to be discussed immediately, other supplementary pieces of information, such as long-term

outcomes, may not be suitable to convey at the same time. Therefore, staff may benefit from telling parents the information is available when they are ready to hear it.

Furthermore, it is no longer only staff who are gatekeepers to information; parents are savvy to seeking out other sources such as websites and printed materials in the hospital, and have opportunities to learn about their child's condition in their own time.

Another key implication for professionals offering emotional support to the family during treatment is that there might be low uptake due to the focus on the child's physical health. The process framework, as seen in Figure 3, would suggest that towards the end of treatment would be more realistic as this is when parents begin to reflect on their experiences.

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Part 3: Critical Appraisal

Reflections on qualitative research within a paediatric brain tumour population

Introduction

This critical appraisal will focus on three areas of the research process, from the earliest stages of design and recruitment to data collection and analysis. I kept a research journal to write down my thoughts, and have used this to guide my appraisal. Firstly, I will reflect on my reasons for working with the paediatric cancer population and the clinical utility of the qualitative design (Conducting Research with Parents of Paediatric Brain Tumour Survivors). I will next consider the trade-off between opting for a homogeneous sample over a heterogeneous sample, how this affected the recruitment process, and the clinical implications of this (The Population Sample and the Recruitment Process). Finally, I will appraise my analysis, and discuss the utility of incorporating quantitative methods for further research (Using Thematic Analysis).

1. Conducting Research with Parents of Paediatric Brain Tumour Survivors

In this section, I think about my reasons for choosing to work with parents of brain tumour survivors and how I handled the emotional content of the interviews. I then consider how the qualitative design enabled the participants to have a voice that was evident in my write-up.

The emotional content of the interviews

Before my doctoral training I worked as an assistant psychologist investigating the psychological outcomes of paediatric brain tumour survivors. Due to time and resources,

the focus was often on the child alone and little attention was paid to the family; to the parents/caregivers, who have been shown in research to suffer anxiety and depressive symptoms as a consequence, as well as to the siblings, who can feel ignored and resentful (Houtzager et al., 2004). One of the most jarring experiences was when a father of a child who had developed profound cognitive and physical disabilities stated that he now had ‘a different child’. I believe he was slowly coming to terms to adjustments within both of their lives but I was surprised that this adjustment had continued for many years post-treatment. My literature review (the first part of this thesis) indicated that there is paucity of research into the distressing and enduring process of caring for a sick child and coping with psychosocial changes. Following this, the findings from my empirical paper included advice for staff and other parents about how to support this process. As written in my research journal, conducting this study allowed me to satisfy the ‘lingering sadness I felt for the father’.

Within my study, all participants expressed that there were adjustments within the family. One of the most surprising findings was how quickly I felt I reached saturation of interviewees’ shared experiences despite the diversity of families in the sample, especially when considering that childhood brain tumours are, by all intents and purposes, random and not selective to specific populations (Patenaude & Kupst, 2005; Upton & Eiser, 2006). For example, post-traumatic growth emerged as a prevailing concept, as a way to re-evaluate life and look for meaning and happiness (O’Leary & Ickovics, 1995; Hefferon, Greal, & Mutrie, 2009). As can be seen in my results section (with the domain ‘Finding a new normal’ and the theme of ‘Hope for the future’) the adjustment process was comparable for many parents. With this in mind, I was

encouraged that many of the parents were able to arrive at a point in life in which they felt optimistic despite the struggles they encountered along the way.

I believe parents' optimism of their current circumstances made the interviews easier to conduct. Although I found many of the interviews upsetting, due to the traumatic experiences and feelings of helplessness during the initial treatment, I did not find the conversations to be as distressing as I first imagined. I think this was because the parents were able to talk about new-found positive appraisals and could look to the future, despite earlier physical and psychosocial setbacks to the child. At the time of the interviews, the children were alive and, on the whole, healthy and I think this helped contain earlier distress parents had experienced; had the interviews, for example, occurred as treatment was ongoing, or had I included families in which the child had died, I think this would have resulted in potentially more distressing discussions.

I was also aware that, as a researcher, I wanted to maintain professionalism, allowing parents to guide the conversation without my own reactions becoming a prominent part of the interview. At the same time, I was concerned that not showing any emotion could seem cold and disinterested, and possibly affect any therapeutic warmth that aided their narratives. I believe the way I managed this was to embrace a person-centred position, of being warm, genuine and empathic (Rogers, 1986) and I saw the value in these core conditions whilst conducting qualitative research.

Enabling the participants to have a voice

One particular issue that I struggled to resolve relates back to the man who fostered my interest in this population and the language he used, stating that he had 'a different

child'. I was intrigued by the choice of words: different to how the child was before, different to other children, or different to what the father expected and hoped for the child? Many of the parents I interviewed used similar terminology: "Even if you have a normal child you can't see what the future holds, because they might not academically achieve things" (P7). Although 'normal' and 'different' and so on were words parents used, I did not feel comfortable writing about parents no longer having 'a normal child', considering that readers may find this insensitive, and I questioned how to include these words in my write-up.

I brought this to supervision and decided on the usefulness of retaining the same phraseology in the write-up. We justified this by grounding the words in examples from the original data as well as aiming to evoke emotion and provoke responses that are meaningful to the reader (Sandelowski, 1994). Whilst some readers may find the terms jarring, giving a voice to the participants is at the heart of the qualitative design and their choice of words should, therefore, be acknowledged (Hunt, 2011).

2. The Population Sample and the Recruitment Process

An early contention with the design was whether the sample should be heterogeneous or homogeneous, in this case stratified by tumour type. I understood that a tighter-knit group would strengthen any patterns that emerged from the analysis. Nevertheless, I saw two distinct disadvantages of using a homogeneous group. Firstly, homogeneity could limit the pool of potential participants and risk an unreliable sample size. Despite this, recruitment was successful. The second concern was that the findings would be confined

to one type of brain tumour yet the clinical implications may have relevance to neuro-oncology more broadly. In this section I reflect on reasons for the successful recruitment uptake from a small participant pool. Following this, I consider the limitations of a homogeneous sample when attempting to broaden the findings to similar populations.

Reflections on the successful recruitment uptake

No numbers are given specifically for sample sizes in thematic analysis. However, in line with previous qualitative projects within this context and discussions with supervisors, it was decided that aiming for fourteen participants would be suitable (Braun & Clarke, 2006). The hospital's clinical research committee requested that we investigate one aggressive tumour type, namely medulloblastoma (Johnson et al., 1994), so that all families involved would have witnessed significant changes to quality of life. In this case, homogeneity resulted in a sharp decrease in potential sample size from over one hundred families being eligible to thirteen. Incidentally, only less intrusive tumour types offered larger numbers than this. My concern was a practical one. With thirteen participants from which to recruit, if any parents opted out, withdrew consent, or the child had a tumour recurrence, I could be risking a sample size too small to be credible. Eventually, ten parents were recruited and interviewed, with the other three being unavailable. This high opt-in rate has been described as remarkable in itself by hospital staff, and they have offered four hypotheses why uptake for participation was successful.

Firstly, the hospital is popular and received positive feedback from all the participants involved: "I appreciate all of the staff and want to say thank you very much.

They have done a fantastic job looking after her a lot. I am grateful for them” (Mrs K). Approval from the hospital might have influenced the participants’ confidence that the research was valuable and credible. Furthermore, staff members from the department have kept in regular contact with the families, both for scheduled check-ups as well as informal checking in. This closeness might have allowed parents to feel links to the hospital and its research, compared to other settings where they might have felt like ex-service users and not wanted to return to the past. This is a very good reflection on the hospital and its staff, and I was sure to highlight this in my dissemination.

Secondly, the research was sold to potential participants as a way for them to give feedback to staff and other parents, as explained in the Participant Information Sheet (Appendix E). Participants might have been motivated by the idea that their opinions and advice could inform clinical practice. This hypothesis suggests that the participants were eager to have their say, as well as indicating the generosity of their time and desire to help others in the future, and supports the reason service-user forums are integral to the running of clinical services (Telford & Faulkner, 2004).

The third hypothesis for excellent uptake in recruitment was related to the design, namely that parents were the participants rather than the children. According to staff, research through the hospital typically focuses on the child’s wellbeing and outcomes. Following treatment, in which the child is removed from school for operations, chemotherapy, radiotherapy and other investigations, not to mention for check-ups during the recovery period, parents are eager to integrate the child back into their social and educational environments (Bjork, Wiebe, & Hallstrom, 2005; Alderfer et al., 2010). This research did not focus on the child as the interviewee, and perhaps parents were

happy that they did not have to remove their child from school or put them under further duress in a new medical context.

The fourth and final explanation concerned the content of the interview schedule; it was the opportunity for parents to reflect on their experiences that made them want to take part (Appendix E). Many parents told me that they didn't feel comfortable discussing the past with other family members or friends, and that often they felt the time with staff should be spent on discussing their children's health. Psychology sessions were offered to parents as standard procedure at the hospital around the time of diagnosis and treatment but it may have been only a few years later that parents felt they were only then ready and willing to talk about the past: "If you'd have asked me a couple of years ago to take part I would have said no. I wasn't ready to look back" (P6). Many participants reported the interview to be a cathartic process and the means to look back and reflect on what they had endured and overcome.

I would conjecture that the reason for successful recruitment was a combination of these reasons, which indicates that conducting similar studies in the future that focus on this population should be feasible. As an addendum, perhaps there is a more practical and simpler explanation. Participants were told that they would be interviewed wherever they felt more comfortable and wherever was less hassle for them: at home or at the hospital. Every interviewee chose to be met at their home. Whether the ease of participation and the briefness of interviews aided recruitment is difficult to establish but it may be a contributing factor.

Broadening the findings from a homogeneous sample

As well as the small sample size, the other reason for being apprehensive about using a homogeneous sample was not being able to generalise the findings to other tumour types. Whilst different tumour types require different treatment outcomes and heterogeneity in cancer can miss specifics (Mancini et al., 2011), research suggests that there is much overlap within neuro-oncology (Thibodeaux & Deatrck, 2007; Long & Marsland, 2011). By associating this research with medulloblastoma alone, I was concerned it would deter parents and professionals from looking at the findings when considering other tumour types.

With the exception of one parent who worked as a nurse for cancer patients, no participant had prior knowledge about brain tumours. When learning from staff, leaflets and online searches, parents were satisfied at the level of 'brain tumour' and none had specific knowledge about how medulloblastoma differed. One mother stated that she had enough to think about without the extra details: "I'd just heard that whoever had a brain tumour didn't live, they don't survive. I didn't know the type but just wanted to know the treatment" (P2). Including all tumour types and noting differences may have allowed for broader inclusion when disseminating the findings but I am appreciative of the usefulness of homogeneity (Bowers, Pharmer, & Salas, 2000). I am quietly confident, however, that the findings of this paper can be useful to all families of children with brain tumours, but further research will be needed to explore if there are differences between tumour types.

On reflection, I am pleased that my findings are credible due to the homogeneity of the group. However, I would be interested in exploring the similarities and differences between children with medulloblastoma and other brain tumour types.

3. Using Thematic Analysis

In this final section of the appraisal I consider my decision for using a qualitative design, and how future research may benefit from mixed-methods. I conclude with an evaluation of my rationale to use a thematic analysis in order to generate meaningful data.

Using a qualitative design

Most of the existing research on parental coping within paediatric neuro-oncology is quantitative. My rationale for a qualitative study was that it would be an appropriate method to explore the participants' inner worlds. For many parents, I believe the interview was the first time the focus was on their subjective experiences; asking them to complete questionnaires and other quantitative outcome measures may not have captured personal reflections. I was glad to have selected this design, and wrote in my journal that the process seemed cathartic for parents. Many parents also expressed their gratitude for the opportunity to talk and that it was a positive experience, and this is something quantitative testing would struggle to encapsulate. For example, a questionnaire may have determined that school involvement was important but nowhere would I have recorded the following anecdote from one mother: "He's one of the most

popular children, I think, at school. He went in six months ago when there was all the snow, and it was slippery in the playground and I was worried because he does still trip and fall. The minute he got through the fence there were boys, one either side of him, with their arm underneath him. I could have cried. It's things like that." (P6).

At the same time, research has shown that there is variability in parents' ability to cope with the stress from the treatment and its after-effects, which can then hinder effective care-giving duties (Hassall, Rose, & McDonald, 2005). Through qualitative methods alone, I was not able to quantifiably measure stress reactions other than by asking participants to subjectively rate how they were coping. Differences in levels of stress may be an important factor when asking parents to talk about their experiences of coping. Therefore, if I were to conduct this study again, I would consider including a mixed-methods design, incorporating the interview and a battery of outcome measures to test psychological wellbeing. This may then offer further insight into any discrepancies between different parents' experiences (Bryman, 2006; Ivankova, Creswell, & Stick, 2006).

The rationale for thematic analysis

My aim of the study was to carry out a process in which to code a series of interviews and, from this, offer a coherent picture of the population's experiences. I also wanted to conduct the interviews with no theory-led position so that new or unexpected themes could emerge bottom-up from reading the data (Boyatzis, 1998), and I was aware that thematic analysis allowed for inductive research. Furthermore, my

understanding of thematic analysis was that it is not bound by any epistemological stance and catered for flexibility.

There are many quality frameworks and checklists for appraising qualitative methods and, by drawing from credible references, I was able to achieve what I believe is a thorough design for a thematic analysis (McLeod, 2001; Spencer et al., 2003; Braun & Clarke, 2006; Pistrang & Barker, 2010). Some of the checks were easier to administer, such as including a table indicating how often themes were mentioned, as well as using quotes in my results section to give my findings validity.

One of the credibility checks I found the most interesting was in making the findings meaningful to the reader and evoking emotional responses (Sandelowski, 1994). Thematic analysis necessitates an interpretation of the encoded data and it was not simply enough to tally emerging ideas and bullet-point them. Instead, I analysed the results under meaningful themes, and placed them within a framework for a process, which flexibility within this analysis allowed for (see Figure 2 on page 58). Doing so, all staff members, families and researchers could read my paper and have a clear visual aid for when themes may present themselves (Kerner, Rimer, & Emmons, 2005; Oerman et al., 2008).

At the same time, phenomenological interpretation, using one's experiences to make sense of another's subjective feedback, becomes a vital part of the thematic analysis (Smith, 1995; Braun & Clarke, 2006). However, during analysis I started to question my method. As I wrote in my research journal, 'why am I doing thematic analysis and not interpretative phenomenological analysis (IPA)?' Thematic analysis and IPA seemed, at least by first glance, to be similar. There appears to be little in the way of qualitative

analysis without subjective interpretation and, in that line of thought, little in the way of interpretation without phenomenology. Furthermore, their philosophies appeared to be in line with one another (Braun & Clarke, 2006; Brocki & Wearden, 2006). Even articles comparing qualitative methods seemed to come undone at this point, although some were better than others at highlighting the differences in the breakdown of stages during analysis (Pistrang and Barker, 2012). I found my answer in supervision, realising that IPA had a criteria for concentrating on much smaller sample sizes (Smith, 2004), whilst thematic analysis was more appropriate for larger samples (Braun & Clarke, 2006).

Overall, I found the process of thematic analysis helpful for coding and organising my themes by domains. Moreover, I was able to realise that themes occurred in a longitudinal process, and decided to present the information accordingly (see Figure 3 on page 70). I believe that this was a successful piece of research, due in a large part to the way the results took form, and I would conjecture that the flexibility thematic analysis allows contributed to this.

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Appendices

Appendix A: Qualitative Review Checklist (adapted from Letts et al., 2007)

1. Is there a clear statement about the aims of the research?
2. Is qualitative methodology appropriate?
3. Is the design appropriate for the purpose of the study?
4. Does sampling and recruitment meet the needs of the study?
5. Did testing continue until saturation was met?
6. Was consent given, and were participants able to opt-out?
7. Are researchers' credentials and assumptions transparent?
8. Are testing methods explained comprehensively?
9. Is the process of analysing the data into themes described?
10. Are results meaningful to the reader?
11. Are results rich with descriptions and consistent with raw data?
12. Are the conclusions appropriate to the study's aims?
13. Do the findings inform theory/clinical practice?

Appendix B: Quantitative Review Checklist (adapted from Downs & Black, 1998)

- 1 Is the hypothesis/aim/objective of the study clearly described?
- 2 Are the main outcomes to be measured clearly described?
- 3 Are the characteristics of the patients included in the study clearly described?
- 4 Are the interventions of interest clearly described?
- 5 Are the distributions of principal confounders in each group of subjects to be compared clearly described?
- 6 Are the main findings of the study clearly described?
- 7 Does the study provide estimates of the random variability in the data for the main outcomes?
- 8 Have adverse events that may be a consequence of the intervention been reported?
- 9 Have the characteristics of patients lost to follow-up been described?
- 10 Have actual probability values been reported (e.g. 0.035 rather than <0.05)?
- 11 Were the subjects asked to participate in the study representative of the entire population from which they were recruited?
- 12 Were those subjects who were prepared to participate representative of the entire population from which they were recruited?
- 13 Were the staff, places, and facilities where the patients were treated, representative of the treatment the majority of patients receive?
- 14 If any of the results of the study were based on “data dredging”, was this made clear?
- 15 Were the statistical tests used to assess the main outcomes appropriate?
- 16 Were the main outcome measures used accurate (valid and reliable)?
- 17 Were losses of patients to follow-up taken into account?
- 18 Did the study have sufficient power to detect a clinically important effect where the probability value for a difference being due to chance $<5\%$?

Appendix C. Hospital's Clinical Research Committee Approval



Joint Research and Development Office
Division of Research and Innovation

Mr Phil Lurie and Dr Dianne Gumley
Clinical Psychology
Great Ormond Street Hospital

3 May 2013

Dear Phil and Dianne

Title: Adjusting to life after a paediatric brain tumour: A thematic analysis of parents' experiences
R&D Ref: 13NR15
Funding: Clinical Own Account
Decision: Approval

Thank you for your detailed responses to the issues raised by the CRAC committee, which I have reviewed on behalf of the committee. I am pleased to confirm CRAC approval for the project.

You will shortly receive a checklist of documents that are required for R&D approval. Once all the documents have been received you will receive an R&D approval email and you can commence your project.

Regards

A black rectangular box redacting the signature of Dr John Anderson.

Dr John Anderson
Chair
Clinical Research Adoption Committee

Joint Research and Development Office
Division of Research and Innovation
UCL Institute of Child Health, 30 Guilford Street, London WC1N 1EH
Tel: 020 7905 2179 Fax: 020 7905 2201
www.gosh.org

The child first and always

Appendix D. NHS Ethics Approval



Health Research Authority

NRES Committee London - Stanmore

Skipton House
Ground Floor
NRES/HRA
80 London Road
London
SE1 6LH

Telephone: 020 7972 2552

20th June 2013

Mr Philip Lurie
UCL Division of Psychology and Language Sciences
1-19 Torrington Place
London
WC1E 7HB

Dear Mr Lurie

Study title: Adjusting to life after a paediatric brain tumour: A thematic analysis of parents' experiences
REC reference: 13/LO/0781
IRAS project ID: 107254

Thank you for your letter of 18th June 2013. I can confirm the REC has received the documents listed below and that these comply with the approval conditions detailed in our letter dated 14th June 2013.

Documents received

The documents received were as follows:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Covering Letter		18 June 2013
Participant Consent Form	5	18 June 2013
Participant Information Sheet	5	18 June 2013
Protocol	3	18 June 2013

Approved documents

The final list of approved documentation for the study is therefore as follows:

<i>Document</i>	<i>Version</i>	<i>Date</i>
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Covering Letter		18 June 2013
Evidence of insurance or indemnity		30 July 2012
Interview Schedules/Topic Guides	Draft Qualitative Interview Schedule	09 May 2013
Investigator CV	Stephen Butler	09 May 2013
Letter from Sponsor		28 November 2012
Other: Student CV: Mr Philip Lurie	1	09 May 2013
Other: Clinical Research Adoption Committee Approval (GOSH)		03 May 2013
Other: Letter from Funder (Email from Oxana Brigden)		22 February 2013
Other: CV: Dr Gumley		
Participant Consent Form	5	25 April 2013
Participant Consent Form	6	18 June 2013
Participant Information Sheet	5	25 April 2013
Participant Information Sheet	6	18 June 2013
Protocol	3	18 June 2013
REC application	107254/447848/1/343	02 May 2013
Summary/Synopsis	Participant Flow Chart	09 May 2013

You should ensure that the sponsor has a copy of the final documentation for the study. It is the sponsor's responsibility to ensure that the documentation is made available to R&D offices at all participating sites.

13/LO/0781	Please quote this number on all correspondence
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Yours sincerely



Ms Julie Kidd
Committee Co-ordinator

E-mail: Juliekidd@nhs.net

Copy to: *Dr Stephen Butler Stephen.butler@ucl.ac.uk*
Mr David Wilson, clara.kalu@uclh.nhs.uk

Appendix E: Participant Information Sheet

UNIVERSITY COLLEGE LONDON



PARTICIPANT INFORMATION SHEET

Title of Project: Coping with a childhood brain tumour: An analysis of parents' experiences

Name of Researcher: Phil Lurie

Introduction

I would like to invite you to take part in my research study. Before you decide, it is important to understand why the research is being done and what it involves.

I am investigating the ways in which parents cope and manage their expectations of their children following treatment of a brain tumour. The findings may provide useful strategies for other parents in the future.

What happens if you agree to take part?

If you agree to participate I will meet with you for about one hour and ask you questions about your experiences. To make it as convenient and comfortable as possible, you may choose whether to meet at a clinic room in Great Ormond Street Hospital or at your home.

I will record our conversations and transcribe them into written text. Following this, I will make the data anonymous, by removing all names and identifying information. You will be able to read over the transcript to verify what was said and I will feed back my findings to you.

Why should I take part?

The benefits to participating are that participants may appreciate the opportunity to reflect on their experiences. Participants will also provide information that may help parents in the future. Due to the sensitive nature of this discussion, participants are advised that they may become upset during the interview. In this event, participants may choose to opt out of the study. Participants who wish to discuss anything further will be offered to meet with a member of the Great Ormond Street Paediatric Psychology team.

If you decide to travel to Great Ormond Street, I am able to reimburse your travel expenses of up to £3 each way. Every participant will receive a £10 gift voucher as a thank you for their time.

Rules I must follow

There are a few things for you to know before you decide whether or not to take part in this study:

1. Consent

You do not have to take part if you do not want to. If you agree to take part you are able to change your mind and withdraw at any time, without giving a reason. This would not affect any standard of care you or your family receive in the future. Should you withdraw, all data collected up to that point will be destroyed.

2. Confidentiality

All information will remain confidential. Your interview will be audio-recorded, on a protected device. Data will be stored in a secure area and not shared with anyone outside the study.

3. Reporting the findings of the study

A report will be written about the results. The results will be presented in such a way that no one can identify you or your family, or know that you took part.

4. Ethical approval for research

All research in the NHS is looked at by a Research Ethics Committee to protect your interests. This research was given a favourable opinion by on 20th June 2013.

What if there is a problem

If you wish to complain, or have any concerns about any aspect of the way you have been approached or treated by members of staff you may have experienced due to your participation in the research, National Health Service or UCL complaints mechanisms are available to you. Please ask your research doctor if you would like more information on this.

In the unlikely event that you are harmed by taking part, compensation may be available.

If you suspect that the harm is the result of University College London or the hospital's negligence then you may be able to claim compensation. After discussing with your research doctor, please make the claim in writing to Dr Stephen Butler who is the Chief Investigator for the research and is based at the address below. The Chief Investigator will then pass the claim to UCL's Insurers. You may have to bear the costs of the legal action initially, and you should consult a lawyer about this.

Patient Advice & Liaison Service

Great Ormond Street Hospital
London
WC1N 3JH
Tel: 0207 7829 7862

Conclusion

I am happy to discuss any aspect of this research with you. What I learn from this research may be beneficial to other families of brain tumour survivors, and I hope you will find it interesting to take part.

Thank you for your time,

Phil Lurie
Trainee Clinical Psychologist

University College London
Department of Clinical Health Psychology
London
WC1E 6BT
Tel: 020 7679 5699

Dr Dianne Gumley
Consultant Clinical Psychologist

Great Ormond Street Hospital
Department of Paediatric Psychology
London
WC1N 3JH
Tel: 020 7405 9200

Appendix F: Consent Form

UNIVERSITY COLLEGE LONDON



Patient Identification Number for this trial:

CONSENT FORM

Title of Project: Coping with a childhood brain tumour: An analysis of parents' experiences

Name of Researcher: Phil Lurie

Please initial all boxes

1. I confirm that I have read and understand the information sheet dated 25/04/2013 (version 5) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.

2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my medical care or legal rights being affected.

3. I understand that relevant sections of my child's medical notes and data collected during the study may be looked at by individuals from UCL, from regulatory authorities or from the NHS Trust, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records.

4. I understand that my interview will be audio-recorded on a secure and protected device, which will be kept on site and only accessed by members of the research team.

5. I agree to take part in the above study.

_____	_____	_____	_____
Name of Participant	Date	Signature	Name of Child
_____	_____	_____	
Name of Person taking consent.	Date	Signature	

Appendix G: Draft Interview Schedule

Whilst interviews will be flexible (and modified in response to efficacy and/or problems of prior ones) a general schedule is drafted:

Opening

Establish Rapport

Introduce myself, my role, the course and the need to conduct research.

Purpose and Motivation

Explain the rationale and pre-existing literature to help normalise the discussion

Explain the aims of the research, how it will benefit participants, and dissemination

Explain how long the interview should take, and can take breaks whenever needed

Consent

Adhere to ethical standards, read/sign consent. Explain the audio-recording, confidentiality, credibility check (offering participants to read transcription for accurate representation), and the right to opt-out without affecting care. Ask if any questions.

Body (start recording)

General Demographic and Family Information

"It would be helpful to understand a bit about your family set-up..."

- 1) At time of diagnosis: N, ages, jobs, school, ethnicity
- 2) Now: N, ages, jobs, school

Child's psychosocial development and experiences pre-diagnosis:

"Thinking back to how your child was before he/she was unwell..."

- 1) *"How was your child doing at school? Did he/she have friends? Did teachers/others provide you with any feedback?"*
- 2) *"How would you describe your child's personality? How was their mood?"*
- 3) *"How was your child's physical health?"*
- 4) *"Did you have any expectations about his/her future? What aspirations/hopes did you have for him/her?"*

General Child Diagnosis and Treatment Information

"If it is alright, we will now move on to the time when your child was diagnosed..."

- 1) *"Could you tell me how your child was diagnosed and the treatment plan?"*
- 2) *"Did you know anything about brain tumours at that time? How did you learn about them following your child's diagnosis?"*
- 3) *"Did you have any thoughts/discussions with professionals/others about your child's intellectual, educational, emotional, behavioural, social and/or health being affected by a brain tumour? Did any professionals discuss possible outcomes? Did you find information about outcomes elsewhere?"*
- 4) *"Did you have any expectations of changes to your child's intellectual, educational, emotional, behavioural, social development and/or health due to the treatment plan?"*

- 5) *“Did aspirations/hopes/fears change due to diagnosis/treatment? If so, how did you manage the uncertainty of the future during treatment?”*
- 6) *“What strategies did you use to cope during this time?”*

General Treatment and Survival Adjustment Information

“It would be helpful to think about the time after treatment. Before we move on, is there anything you would like to add?”

- 1) *“Did anything at time of completion of treatment for a brain tumour alter your aspirations, hopes and fears for your child?”*
- 2) *“Did you notice any intellectual, educational, emotional, behavioural, social and/or health changes at time of treatment?”*
- 3) *“If so, did this alter your expectations and aspirations of your child?”*
- 4) *“Did you notice any intellectual, educational, emotional, behavioural, social and/or health changes since completion of treatment?”*
- 5) *“If so, did this alter your expectations and aspirations of your child?”*
- 6) *“How did you cope during this time?”*

Advice for New Parents and Health Professionals

“I will finish this interview by asking a few questions about advice that you were given, as well as advice you would give to other parents.”

- 1) *“Were you given any advice/recommendations/help at time of diagnosis/treatment for coping and managing expectations for potential cognitive, educational, emotional, behavioural, social and/or health changes as a result of treatment? If so, when and by whom?”*
- 2) *“Was there any advice/recommendations/help you wish you had/had not been given?”*
- 3) *“What advice would you give to other parents entering the system in the future for coping and managing expectations at time of diagnosis?”*

Closing

Recap what has been said. Ask if anything important hasn't been discussed, or anything participants would like to add/clarify.

Maintaining Rapport and Reimbursement

Reimburse participants with travel money and gift voucher. Ask how they found the experience and whether reflection has been beneficial. Offer them support through GOSH should they wish to take it.

Action to be Taken

Ask if it is alright to contact them should any questions arise. Offer participants the chance to receive their transcription to check for anonymity and check they are happy with the content. Thank them for their time, and turn off audio-recording.

Appendix H: Example of an Analysed Transcript (P7)

don't talk to others who don't understand people are cur.ous

G I'm comfortable talking about it. I don't talk too much about it. Someone might ask about the hearing loss and I might say it happened some years ago and if they probe me I say it was chemo and I leave it at that. I don't want to go into detail - I keep it vague. It could be chemo for anything. They want to know why she has hearing loss. But I only do it when people probe.

OTHERS DON'T UNDERSTAND

How about you thinking about the past, cause a lot has changed in three years?

be happy don't dwell on the past

G It has, but I see it as the past and we're working on it. I push it aside. It's not worth it. As long as she's alive and well and happy, the past is gone and I'm glad it's gone. I don't want to dwell on it but I do worry a bout the tumour recurring. I tend to not think about it but then I get to the MRI scan and the results day. I get anxious thinking what they'll say. I just leave it, I don't see nay point thinking about it for 4/6 months cause I can't function so I put it to the back of my mind and I get to that day and start thinking why things are taking long. Sometimes I wait two hours and then I think I'll get bad news, and it's just a backlog but I get anxious. And I'm reading all these signs, which probably aren't there. If the results are good then I'm happy but then sometimes I worry that I'm not cautious enough. I think she wants to know as well. She's at that age, she knows what happened but doesn't want to talk about it. I tell her I'd rather be honest so if she wants to talk.

VALUES HAVE CHANGED

worry about recurrence link with anxiety honesty. how much to disclose include child

able to put it to back of mind but...never ending, scars continue

Sounds sensible.

G Yeah, I wont tell her fibs, like it's a happy day out.

I'll finish off asking about advice. Were you given any advice/recommendations about what could happen in the future?

every child has different outcomes, and hard to know which until later advice. pursue support lots of education time so important to link to that

G We were given a leaflet about the effects of RT and at the time we were thinking, well, we'll just see what happens, cause it's very individual. Every child has different effects. Hearing loss only affects a few, it seemed to affect her, so yeah. We saw the endocrinologist and she told us to go for a statement, which was good advice and she got it, which was helpful as she got extra time in exams and 20 minutes a day extra with a teacher, and it really helped. It's good to listen to the advice and pursue it, which helps since they spend most of their time in education. I can't think of anything else.

MY CHILD IS NOT THE SAME AS ANOTHER

KEEP SCHOOL INPUT GOING

How about managing expectations of changes. Did anyone tell you about things that might happen that you should consider?

G I don't think so.

Was there anything you wish you'd been told?

wanted more realistic results

G I think I wish I'd been told how much she'd slow down, especially in learning. She talks quickly, she's verbal, but it's just the learning has slowed down a lot. I expected her to be faster than me. I'm older! Even physically she's quite slow. I try to keep her physically active, with swimming and tennis, but she's still finding it hard to keep up.

If you'd been told that at the time, would that have taught you anything?

maybe more prepared

G Maybe not, I might have been more prepared. Compared to other children she is physically slower, and slower at learning things.

Was there any advice you wish you hadn't been given?

better to receive all advice but includes bad advice would be wise to take it in?

G Not really, cause you take it or leave it. If someone gives you bad advice you don't listen. I was told certain things, like medicine. There was an issue with a peg and I was given different advice, prescribed different things. A nurse told me I could use j-cloths as wipes and my community nurse told me you shouldn't be using that. I was told wrong advice by professionals, but I don't know if that was, obviously in the end you can see through bad advice.

BE OPEN TO ALL ADVICE BUT BE CHOOSY

If I could ask you if the hospital phoned and said there were some worries about future expectations of their child, what would you tell the parents?

advice: every child is different
advice: take help, you need breaks

G I'd say every child is different and you can't really compare yourself to another child even if they have the same condition, and then to take help if anyone offers help such as a hospice, you just need a break, just to take it. Sometimes you do need a break, cause the kid's playing up and they're fed up looking at you all day and fed up off the treatment, so someone fresh caring for them can help cause they'll appreciate you more when you're doing it. I think she saw me as the bad mum cause I was making her take medicine and having her feeds, but she said she hated me then, that I was horrible. I said I was trying to help you but you couldn't see that cause I was being mean to you, but that's what I had to do. I used sticker charts to reward good behaviour. I kind of used everything I could, and some of the mums offered to take her out for a bit of the day and that was good. That was some of the mums at the school. They were part of the church and they decided they'd knock on my door and take her to the shops and have an ice cream, and you can lie down.

EVERY CHILD IS DIFFERENT

parent = caregiver doing horrible things which can ruin relationship
practical breaks from other mums

TAKE BREAKS

What if they asked you how to manage concerns about changes in the future?

anything can happen take it day at a time
* don't work off child but don't pressure them

G Well, anything could happen in the future. I don't think there's any way to manage it. You have to take it a day at a time. Your child could, well not get better, but they could have other talents that you can't yet see. Even if you have a normal child you can't see what the future holds, cause they might not academically achieve things. You can't really say 'I want this for my child' cause it might not happen. I'm not sure I'm saying the right things?

ONE DAY AT A TIME

CHILD CAN HAVE OTHER TALENTS - OPTIMISM?

Appendix I: Breakdown of Domains, Themes and Subthemes

