Parental Wellbeing following Paediatric Brain Tumour and Traumatic Brain Injury

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Thesis Portfolio Abstract

Aims: The thesis portfolio aimed to examine distress in parents of children with traumatic brain injury and parents of children with a brain tumour.

Design: The thesis portfolio consists of two main papers and several additional chapters. The first paper was a systematic review that examined emotional distress in parents of children with traumatic brain injury. The second paper was an empirical study that examined post-traumatic stress symptoms in parents of children with a brain tumour. The additional chapters included further information and provided an overall critique of the thesis portfolio.

Results: The systematic review indicated that parents of children with traumatic brain injury are more likely to experience emotional distress; this can be impacted by several factors, such as factors related to parents' and children's characteristics. The empirical paper suggested that a large proportion of parents of children with a brain tumour experience post-traumatic stress symptoms and indicated that disengaged coping may increase the risk of post-traumatic stress symptoms.

Conclusions: The thesis portfolio demonstrated that parents of children with traumatic brain injury and parents of children with a brain tumour are at risk of emotional distress, even years after the initial event or diagnosis. The results suggest that the mental health of these parents should be considered in paediatric services and these parents should be provided with more support where necessary. Future research should explore parental mental health further with longitudinal designs and evaluate clinical interventions for parents affected by paediatric illness.

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Firstly, I would like to thank all the parents who took part; it was a privilege to be able to conduct my research with you and I am extremely grateful for your help with this research project. I would also like to thank staff members whom very kindly helped me to recruit; the project could not have happened without you and I am grateful that you invested your time in the study. I would like to thank Natalie Brunsdun, Assistant Psychologist, for conducting the independent quality ratings for the studies reviewed in the systematic review. I would also like to thank Dave Peck and Briony Gee for your invaluable statistics advice throughout the project. I would like to thank my primary supervisor, Kiki Mastroyannopoulou, and my secondary supervisor, Judith Young, for your support throughout the project and helpful advice. In addition, I would like to thank Angela Kirby, the clinical collaborator, who helped design and implement the project. Finally, I would like to thank my family and friends for your support throughout the process; I am particularly grateful for the support and encouragement from my sister, Hope, and my partner, Aaron.

Introduction to the Thesis Portfolio

The broader aim of this thesis portfolio is to examine the emotional experiences of parents of children who have experienced a potentially life-threatening medical event. The systematic review will examine distress in parents of children with traumatic brain injury (TBI) and the empirical paper will investigate post-traumatic stress symptoms in parents of children with a brain tumour.

Key Terms

Traumatic brain injury (TBI). TBI is an injury to the brain (Headway, n.d.) from an external force (Maas, Stocchetti, & Bullock, 2008). A diagnosis of a TBI is made based on clinical symptoms, such as loss of consciousness (Maas et al., 2008). TBI's can vary greatly between people (Maas et al., 2008) and can contribute to a number of long-term consequences for the individual, such as impacting cognitive functioning (Yaetes et al., 2002) and behaviour (McKinlay et al., 2014).

Brain tumours. Brain tumours refer to a collection of cells in the brain that grow in uncontrolled ways (NHS, n.d.). There are approximately 130 different types of brain tumours (Cancer Research UK, n.d.). Brain tumours can be primary (originate in the brain) or secondary (spread from elsewhere in the body) and can be malignant (referred to as high grade) or benign (referred to as low grade; Cancer Research UK, n.d.). The survival rate for brain tumours varies significantly depending on factors such as the type and location of brain tumour and response to treatment (The Brain Tumour Charity, n.d.). There are various treatments for brain tumours, such as surgery, chemotherapy and radiotherapy (NHS, n.d.).

Post-traumatic stress disorder. Post-traumatic stress disorder (PTSD) is described by the ICD-10 as a response to traumatic events of a threatening or catastrophic nature (World Health Organisation, 1992). In the 4th edition of the DSM (The Diagnostic

and Statistical Manual of Mental Disorders) it was stipulated that PTSD can be triggered by being told one's child has a life-threatening condition (American Psychiatric Association, 1994) and the DSM-5 indicates that the trauma can be experienced indirectly (American Psychiatric Association, 2013). There are several symptoms of PTSD; key symptoms include re-experiencing the event, avoiding or a preference to avoid reminders of the event and hyperarousal (National Collaborating Centre for Mental Health, 2005). There are debates relating to whether cancer-related distress can be explored within a PTSD model (Kangas, Henry, & Bryant, 2002) and, instead, researchers have used the term cancer-related post-traumatic stress symptoms (PTSS) to describe traumatic reactions in these parents (Bruce, Gumley, Isham, Fearon, & Phipps, 2011; Kangas et al., 2002); this term will be used in the empirical paper.

Rationale

Rationale for the empirical paper. The general topic area of the thesis was first discussed by the primary supervisor and the clinical collaborator; both clinicians are psychologists who have worked with parents of children with a brain tumour. The evidence base, to date, was explored by the primary author, and the two recent reviews in this area were consulted (Bruce, 2006; Yalug, Tufan, Doksat, & Yaluğ, 2011). Following these discussions, and a review of the current literature, it was proposed that the relationship between PTSS and several key variables would be explored in parents of children with a brain tumour. These factors had not been researched in this specific population when exploring PTSS and based on clinical observations, the literature and models to date, it was anticipated that these variables could have a significant impact on PTSS.

Rational for the systematic review. Systematic reviews examining the literature regarding PTSS/PTSD in parents of children with cancer have been conducted by Bruce

(2006) and Yalug et al. (2011); these reviews provide a thorough overview of factors related to PTSS in parents of children with cancer. Studies published since this time are largely in keeping with the findings from these reviews and are discussed within the thesis portfolio. To enable a broader understanding of research in this area, the wider evidence base was consulted. Subsequently it was proposed that the systematic review within the thesis portfolio would focus on the mental health of parents of children with TBI. These parents may have similar experiences to parents of children with brain tumours, such as experiencing threat to their child's life and adapting to potential cognitive and behavioural difficulties in their children (Clark, Prior, & Kinsella, 2002; Mckinlay et al., 2014; Moore, 2005; Prasad, Swank, & Ewing-Cobbs, 2017; Yeates et al., 2002). Therefore, due to the similar experiences that parents may share, it was anticipated that research in this area would also be relevant to the empirical paper and would add to wider research on the impact of paediatric illness on parents.

Outline of Thesis

The thesis begins with a systematic review of psychological distress in parents of children with TBI. Following this, there is a bridging chapter which summarises the results from the systematic review and indicates how the review relates to the wider research around parental mental health following paediatric illness. The thesis portfolio then leads on to the empirical paper, which sets out to explore post-traumatic stress symptoms in parents of children with a brain tumour. The empirical paper is followed by an extended methodology chapter and an additional results chapter; these chapters include information that could not be fully detailed in the former chapters. Finally, the thesis portfolio concludes with a critical discussion chapter; this provides an overall summary of all chapters and outlines the main implications, as well as the strengths and limitations of the thesis portfolio.

Chapter 1. Systematic review

Prepared for submission to the 'Journal of the International Neuropsychological Society'*

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*Author guidelines can be seen in Appendix A.

Psychological distress in parents of children with traumatic brain injury: A Systematic Review

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Abstract

Objective: Paediatric traumatic brain injury (TBI) can have a significant impact on parents, with research suggesting that parents can experience high levels of stress and burden. This systematic review set out to examine research that has compared the distress of parents of children with TBI, to a control group. The review also aimed to examine which factors impact on parents' levels of distress.

Method: A systematic search of four databases was conducted. Other articles were also identified through examining reference lists and searching terms in key journals. Articles were included if they compared parental distress in parents of children with TBI with a control group or examined factors that impact parents' distress.

Results: Overall, twenty-four studies were included. More than half of the studies reviewed performed at least one analysis that identified that parents of children with TBI have elevated levels of distress compared to other parents. A range of factors were found to impact parental distress, such as the severity of the child's injury and parents' previous mental health history.

Conclusions: Parents of children with TBI are more likely to experience emotional distress, particularly parents of children with a severe TBI. There are several factors that can increase parents' risk of distress; such as factors related to parents' and children's characteristics. Clinical services need to be aware of the impact of paediatric TBI on parents and provide adequate support for these families.

Keywords: Brain injuries; Caregiver; Child; Mental Health; Outcomes Assessment; Stress.

Introduction

Traumatic brain injury (TBI) is defined as an insult to the brain from an external force (Medscape, 2017). In the UK it has been estimated that 280 children, per 100,000, are admitted to hospital for a TBI (Hawley, Ward, Long, Owen, & Magnay, 2003). TBI can vary in severity and can be classified as mild, moderate or severe (Ghajar, 2000); it is the most common source of disability in young people (Ghajar, 2000). Paediatric TBI has a chronic sequalae and can contribute to poorer neuropsychological functioning (Yaetes, Taylor, Wade, et al., 2002), poorer social outcomes (Yeates et al., 2004), poorer academic skills (Prasad, Swank, & Ewing-Cobbs, 2017; Taylor et al., 2002) and more behavioural problems (McKinlay et al., 2014; Taylor, et al., 2002). Whilst many studies have reported outcomes in relation to moderate-severe brain injury, the potential long-term impact of mild TBI has also been documented (McKinlay, Grace, Horwood, Fergusson, & MacFarlane, 2009; Mckinley, Grace, Horwood, Fergusson, & MacFarlane, 2010).

The impact of paediatric TBI is extensive, extending beyond the child into the family (Max et al., 1998; Rashid et al., 2014). Parents report that the process of caring for a child with TBI can be an emotional experience (Brown, Whittingham, Sofronoff, & Boyd, 2013). Research has reported that family dynamics are negatively impacted (Rashid et al., 2014) and parents experience high levels of burden (Aitken et al., 2009). Qualitative studies have explored parents' experiences further and have reported on parental experiences of fear, anger, self-blame, loss and hopelessness (Aitken, Mele, & Barrett, 2004; Brown et al., 2013; Du Toit, Coetzee, & Beeton, 2013; Foster, Young, Mitchell, Van, & Curtis, 2017; Kirk, Fallon, Fraser, Robinson, & Vassallo, 2015).

In understanding the experiences noted above, it is important to be aware of the specific challenges faced by these parents. Rivera et al. (1996) report that there are unique stressors to parents of children with TBI, beyond the initial medical trauma. TBI can

contribute to significant challenges for families that can impact on caregivers, such as adapting to neurobehavioral difficulties (Wells, Dywan, & Dumas, 2005) and changes in personality (Degeneffe, 2001). Parents have reported stress related to attending to the needs of a child with TBI (Aitken et al., 2004), changed academic aspirations for their child (Khan, Baguley, & Cameron, 2003) and difficulty predicting the future (Savage, DePompei, Tyler, & Lash, 2005).

In understanding the wellbeing of these parents, it is beneficial to draw on models of caregiver coping and wellbeing. Wallander et al. (1989) and Wallander and Varni (1998) propose a model which points to a range of risk and resistance factors that impact on caregivers' adjustment to chronic health conditions. In this model, risk factors can include those related to the child's diagnosis, such as cognitive functioning and the child's functional independence. Resistance factors are also considered, such as the family environment and parental social support. These factors can influence cognitive appraisal and coping strategies, which can then influence parents' mental health. This model suggests that there could be a range of factors that might impact on the wellbeing of parents of children with TBI.

The impact of parents' psychological wellbeing needs to be understood in the context of the wider family. Research examining paediatric TBI has reported associations between children's and parents' distress (Peterson et al., 2013) and research suggests that parental mental health can impact on children's outcomes following paediatric brain injury (Catroppa et al., 2017; Treble-Barna et al., 2016). This research demonstrates the importance of understanding parents' wellbeing to facilitate the development and implementation of targeted psychosocial interventions for families impacted by TBI.

Considering the research above, it is paramount that research assesses the impact of paediatric TBI on parents. In the adult literature, systematic reviews have been

conducted on caregiver emotional distress (Sander, Maestas, Clark, & Havins, 2013) and mental health (Ennis, Rosenbloom, Canzian, & Topolovec-Vranic, 2013) however, to date, the literature around the mental health of parents of children with TBI has not been reviewed. A systematic review on family functioning after paediatric TBI has been conducted (Rashid et al., 2014), but this did not include a specific focus on parental distress. Therefore, a synthesis of the literature on parental distress would be helpful in understanding the extent of psychological difficulties in this population.

In addition to understanding if parents of children with TBI are at a higher risk of psychological difficulties, it would be valuable to review which factors might impact on parents' psychological distress. As noted above, research examining family adjustment in paediatric TBI has highlighted several factors that may be important to address. An understanding of which factors can predict distress has been examined within the adult TBI literature (Sander et al., 2013), but not within the paediatric TBI literature. A more comprehensive understanding of which factors impact on distress could guide clinical interventions in the future.

In light of the research above, this review aims to assess two questions:

1) Do parents of children with TBI experience elevated levels of psychological distress compared to parents of other children?

2) Which factors significantly impact the psychological distress of parents of children with TBI?

Method

The author used guidelines to guide the systematic search and review (Centre for Reviews and Dissemination, 2009; Denison et al., 2013; Moher, Liberati, Tetzlaff, Altman, & Prisma Group, 2009) and referred to similar systematic reviews in this area to

help guide the search process (Ennis et al., 2013; Sander et al., 2013; Shudy et al., 2006). The search was conducted in March-May 2018 and a refresh search took place in August 2018. The Cochrane database and Prospero were searched to check that there were no reviews in progress, or published, in this area. This review was not registered on Prospero.

Eligibility Criteria

As recommended by published guidelines (Centre for Reviews and Dissemination, 2009; Denison et al., 2013), the eligibility criteria were developed in line with the review questions and were structured by PICOS criteria (Population, Interventions, Comparator Group, Outcomes, Study design; O'Connor, Green, & Higgins, 2008); however the category of "interventions" was not used as this was not applicable to the review.

Population.

Inclusion criteria. Articles were included that examined parents of children and adolescents (0-18 years) with a TBI. All TBI severities were included (mild to severe); this aligns with a similar systematic review assessing the impact of TBI (Sander et al., 2013).

Exclusion criteria. Parents of children with Acquired Brain Injuries (ABI) were excluded to reduce heterogeneity in the review, as in similar reviews (Rashid et al., 2014; Sander et al., 2013). Articles that were not written in English were excluded, due to no access to translation services.

Study outcomes.

Inclusion criteria. The review included studies that used a quantitative measure of parental mental health or emotional/psychological distress.

Exclusion criteria. Measures that examined stressors more generally, or outcomes relating to functioning or burden, were excluded; this is consistent with a similar review in

adults with TBI (Sander et al., 2013). The review excluded studies that only used a parental distress measure as an independent variable.

Comparator groups and analyses.

Inclusion criteria. In relation to the first review question, studies were only included if comparing the significance of distress in parents of children with TBI to parents of children without TBI. In relation to the second review question, studies included were required to use statistical testing to explicitly assess factors that impact on distress or explore factors that contribute to differences in parental distress, within a sample of parents of children with TBI.

Exclusion criteria. Similar to a systematic review in adult TBI (Sander et al., 2013), studies were excluded if they did not analyse data from parents of children with TBI separately to control group data. Regarding the second review question, correlational designs assessing relationships were excluded, as in keeping with Sander et al. (2013), as the authors were interested in the difference and/or variance that independent variables could explain, rather than the strength of an association.

Study design.

Inclusion criteria. The review included studies which used any quantitative design. It was anticipated that most designs would be observational, however separate baseline data from intervention studies were included if applicable to the review questions.

Exclusion criteria. Studies that used qualitative analyses only were excluded.

Search Strategy

EBSCOhost was used to conduct the search in March-May 2018 and combined the following databases: CINAHL Complete (Cumulative Index of Nursing and Allied Health

Literature; 1937-2018), MEDLINE (Medical Literature Analysis and Retrieval System Online; 1946-2018), PsycINFO (1887-2018), and PsycARTICLES (1894-2018). A search of MESH terms was conducted and relevant keywords were used. The search used the following search terms: ("TBI" OR "brain injur*" or "traumatic brain injur* OR "head injur*") AND ("parents" OR "mothers" OR "fathers" OR "caregivers" OR "family" OR "mum*" OR "mom*" OR "dad*") AND ("impact" OR "stress" OR "distress" OR "psychological distress" OR "emotional distress" OR "emotional outcomes" OR "wellbeing" OR "mental health" OR "mental illness" OR "anxiety" OR "depression" OR "depress*") AND ("paediatric" OR "pediatric " OR "child* "OR "youth*" OR "young person" OR "adolescent*" OR "teenager*"). These terms were searched in: subject, keyword, title and abstract, so that the search was as inclusive as possible. The search was limited to peer-reviewed articles, dissertations and theses. Articles had to utilise a human population and needed to be written in English. Review articles, conference abstracts and book chapters were excluded.

The reference lists of articles that met criteria were screened to identify any additional articles. In addition, a search was performed on three relevant journals in May 2018: the Journal of Pediatric Psychology (1976-2018); the Journal of Head Trauma Rehabilitation (1986-2018); and the Journal of the International Neuropsychological Society (1995-2018). To decrease publication bias, the "grey literature" was searched (as in similar reviews; Rashid et al., 2014); to enable this dissertations and theses were included in the search and key authors were contacted about any unpublished papers they had.

Data Analysis

It was expected that studies included in the review would be too heterogeneous to perform a meta-analysis due to using a mix of parents of children with different ages,

different injury severities and assessing parents of children at different time points post-injury. Therefore, it was decided that a narrative synthesis was appropriate; this is consistent with reviews in this area (Ennis et al., 2013; Rashid et al., 2014; Sander et al., 2013). The narrative synthesis was guided by published guidelines on conducting and reporting narrative syntheses (Centre for Reviews and Dissemination, 2009; Popay et al., 2006).

The main findings of the studies were separated into questions one and two to answer the review questions. The findings were analysed by providing a preliminary synthesis of results and then exploring between and within-group similarities and differences, as recommended by Higgens and Green (2011). Several studies related to review question one used more than one analysis to examine differences in distress between parents of children with TBI and control groups. Consequently, for this question, the studies were reviewed as a whole and studies that found at least one significant result were compared to studies that didn't find any significant results. In examining review question two, the focus was on analyses that were significant.

Data Extraction

For each study, the following information was extracted: participant characteristics, study design, time points assessed, study measures and main findings. The main author performed the first data extraction and a second reviewer checked this to ensure accuracy.

Quality of Studies and Risk Bias

A quality assessment was used in this review, however there is no "gold standard" for use in observational studies (Lang & Kleijnen, 2010). This review used an adapted version of the Downs and Blacks scale (Downs & Black, 1998). This scale has been used

in a similar review (Ennis et al., 2013) and has shown to have good test-retest reliability, inter-rater reliability and criterion validity (Downs & Black, 1998). Similar to other studies that have utilised the checklist (Atkinson et al., 2016), this review modified the scale so that it was appropriate to the studies included. In adapting this scale, the STROBE guidelines (Von Elm et al., 2007) and standard assessment quality criteria guidelines (Kmet, Lee, & Cook, 2004) were consulted and a second reviewer was asked to check the applicability of the adapted version by piloting it.

The quality criteria were broadly separated into reporting quality and methodological quality. This study used the qualitative descriptor categories described by Hooper, Jutai, Strong, and Russell-Minda (2008; "excellent", "good", "fair" and "poor") and adapted the scoring to the reduced number of items (excellent 11-12; good 9-10; fair 7-8; poor <6). The review also reported the overall raw score as an indicator of study quality, as used by Ennis et al. (2013). The overall quality was considered in the interpretation of studies. The two reviewers independently rated the quality of every paper and these were compared, using Cohen's Kappa (a measure of agreement; Cohen, 1960), and any discrepancies in ratings were discussed and resolved. The Cochrane risk of bias tool (Higgens et al., 2011) was not used in this review because the tool was not appropriate for the observational studies reviewed. Nevertheless, the Downs and Black (1998) checklist included questions related to bias, such as examining missing data.

Results

Study Inclusion

As seen in Figure 1, four databases were searched using the key terms; this initially yielded 1,930 articles. Other searches were conducted to identify articles, including contacting researchers in the field for unpublished work and searching in three key journals (as noted above); this identified eight articles (all found in key journals).

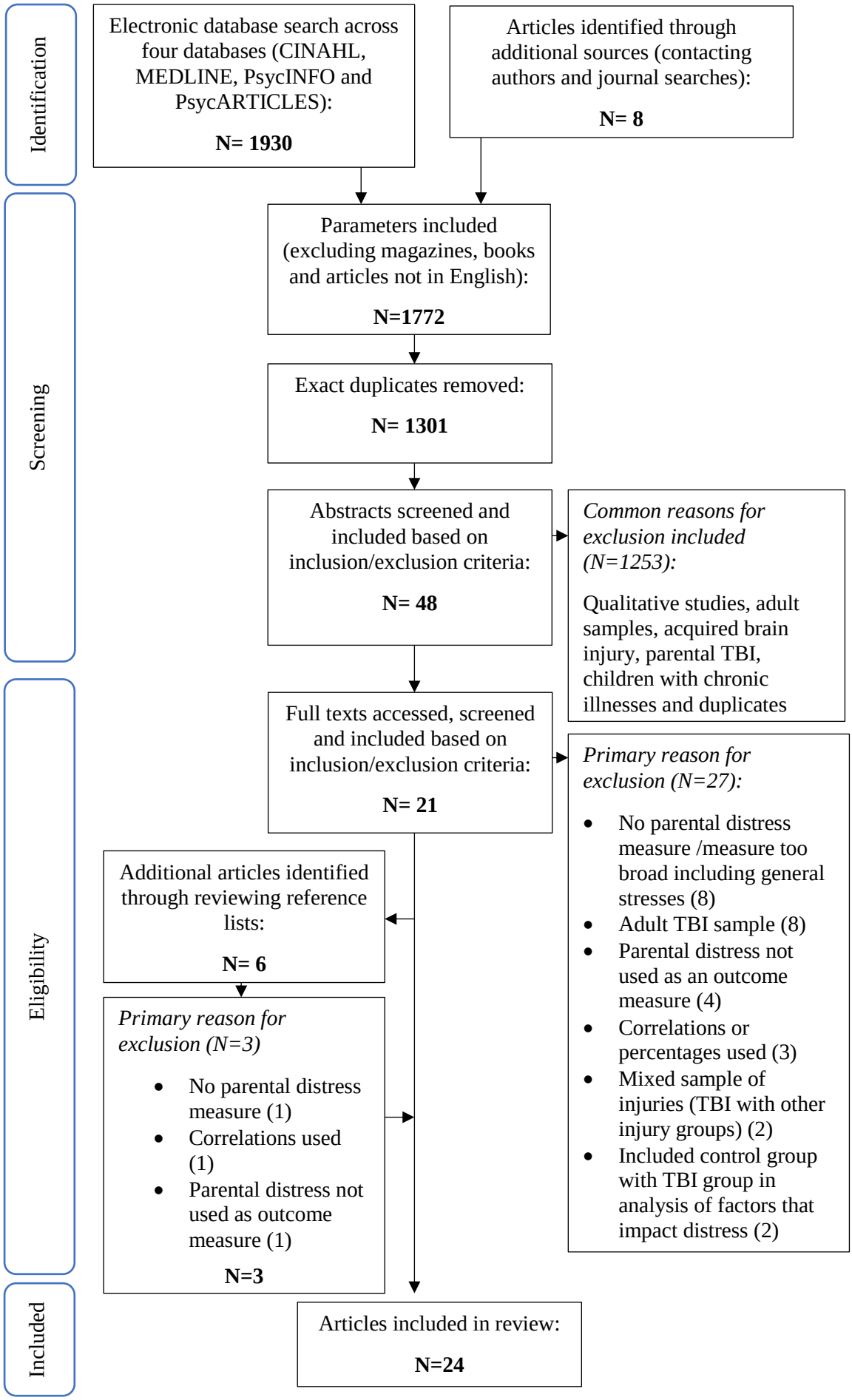


Figure 1. A flowchart outlining the search process. Adapted from "Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement," by D. Moher, A. Liberati, J. Tetzlaff, and D.G Altman, The PRISMA Group, 2009, PLoS Med 6(7): e1000097.

After these identification searches had been conducted, parameters were included, and duplicates removed, resulting in 1,301 articles. Abstracts were screened, and 48 articles met the inclusion/exclusion criteria. Full copies of these articles were requested and 21 were included. A reference list search was then completed for all articles that had been screened fully and three relevant articles were found. Overall, 24 articles met the final inclusion/exclusion criteria. A refresh search took place in August 2018 and no more relevant articles were found.

Quality of Studies

The quality of each study was analysed, using an adapted Downs and Black checklist (Downs & Black, 1998). A second reviewer did an independent rating of quality. Cohen's Kappa was .64 ($\kappa = .64$, $p < .001$); this score indicates moderate agreement (McHugh, 2012). Individual scores for every question were assessed and both reviewers discussed differences and agreed on the overall score. A summary of the quality scores can be seen in Table 1. Overall, studies scored a quality rating of between fair and excellent; no study scored as poor and, therefore, none were excluded from the analysis. The majority of the studies received a rating of good. On average, studies scored better on reporting quality than methodological quality. Common weaknesses across studies included not acknowledging/dealing with missing data, not controlling for confounding variables and not reporting exact significance values.

Table 1

Quality review of studies

Study	Review question answered	Reporting quality (Max: 6)	Methodological quality (Max: 6)	Overall score (Max: 12)	Descriptor
Anderson et al. (2013)	Question 1	6	4	10	Good
Durber et al. (2017)	Question 1	6	4	10	Good
Durish et al. (2017)	Question 1	6	4	10	Good
Ganesalingam et al. (2008)	Question 1	5	5	10	Good
Goldstrohn and Arffa (2005)	Question 1	4	5	9	Good
Hobart-Porter et al. (2015)	Question 2	5	4	9	Good
Micklewright et al. (2012)	Question 1	6	5	11	Excellent
Narad et al. (2016)	Question 1 and 2	6	5	11	Excellent
Raj et al. (2013)	Question 2	5	5	10	Good
Rivara et al. (1996)	Question 2	4	3	7	Fair
Ryan et al. (2016)	Question 1	6	4	10	Good
Stancin et al. (1998)	Question 1	5	5	10	Good
Stancin et al. (2008)	Question 1 and 2	5	5	10	Good
Stancin et al. (2010)	Question 1	5	5	10	Good
Taylor et al. (2001)	Question 1 and 2	4	6	10	Good
Wade et al. (1996)	Question 1 and 2	5	5	10	Good
Wade et al. (1998)	Question 1	5	5	10	Good
Wade et al. (2001)	Question 1 and 2	5	5	10	Good
Wade et al. (2002)	Question 1	4	5	9	Good
Wade et al. (2005)	Question 1	5	6	11	Excellent
Wade et al. (2010)	Question 2	4	5	9	Good
Yeates, Taylor, Woodrome, et al. (2002)	Question 1	5	5	10	Good
Youngblut & Brooten (2006)	Question 2	4	4	8	Fair
Youngblut & Brooten (2008)	Question 2	5	4	9	Good

Risk of Bias

As indicated above, risk of bias was partly assessed through the quality criteria. Some of the methodological weaknesses present, as indicated through the quality review, may have increased bias in some of the studies. The author included dissertations, theses and unpublished work in the review search to reduce possible publication bias. However, no theses or dissertations met study criteria and no unpublished work was found, possibly leading to an increased chance of publication bias.

In the final papers reviewed it was evident that seven studies came from one cohort (Taylor et al., 2001; Wade et al., 2001; Wade et al., 2002, Wade et al., 2005; Wade, Taylor, Drotar, Stancin, & Yeates, 1996, 1998; Yeates, Taylor, Woodrome, et al., 2002), six studies from another cohort (Durber, Yeates, Taylor, Stancin, & Wade 2017; Durish et al., 2017; Narad, Yeates, Taylor, Stancin, & Wade, 2016; Stancin, Wade, Walz, Yeates, & Taylor, 2008, 2010) and two studies from another cohort (Anderson et al., 2013; Ryan et al., 2016). No study duplicated the same statistic/analysis and, therefore, none were excluded but the use of overlapping samples may add bias to the research field as a whole.

Study Details

Main study details can be seen in Table 2.

Table 2

Characteristics of studies included in the review

Authors	Review question/s answered	Sample and participant characteristics	Study design	Study measure/s	Main findings for distress in parents of children with TBI compared to control groups (significance where $p < .05$)	Main findings for significant factors that impact distress in TBI group (significance where $p < .05$)
Anderson et al. (2013)	Question 1	**TBI group/s:** Mild, moderate and severe (N= 93) **Control group/s:** **"**Healthy" children (N=43) **Age range of sample at recruitment:** 5-15 years	**Design:** Cross-sectional design **Time points assessed:** One-time point (at time of injury)	**Measure/s:** GHQ	**Analysis:** ANOVA **Finding 1:** No differences in mental health between groups ($p = .465$)	
Durber et al. (2017)	Question 1	**TBI group/s:** Moderate and severe (N=54) **Control group/s:** OI (N=70) **Age range of sample at recruitment:** 3-7 years	**Design:** Prospective cohort design **Time points assessed:** 5 weeks post injury and 6.83 years after this	**Measure/s:** BSI-GSI	**Analyses:** ANOVA's **Finding 1:** Significantly more distress in severe TBI group (compared to OI group) at 5 weeks ($p = .03$). **Finding 2:** Significantly more distress in severe TBI group (compared to OI group) at 6.83 years ($p = .041$).	
Durish et al. (2017)	Question 1	**TBI group/s:** Severe and mild/moderate (N=60) **Control group/s:** OI (N= 74) **Age range of sample at recruitment:** 3-6 years	**Design:** Prospective cohort design **Time points assessed:** Initial baseline assessment	**Measure/s:** BSI-GSI for baseline assessment and SCL-90-R follow-up	**Analyses:** ANOVA's **Finding 1:** Significant differences in distress between severe and moderate TBI group and	

			and final follow up (average 6.8 years since injury)		OI group at initial assessment (p = .03) **Finding 2:** Significant differences in distress between severe and moderate TBI group and OI group at late follow-up (p = .03)	
Ganesalingam et al. (2008)	Question 1	**TBI group/s:** Mild (N=181) **Control group/s:** OI (N= 97) **Age range of sample at recruitment:** 8-15 years	**Design:** Prospective design **Time points assessed:** 2 weeks and 3 months post-injury	**Measure/s:** BSI-GSI	**Analysis:** MANCOVA **Finding 1:** No differences in distress between groups (p > .05)	
Goldstrohm and Arffa (2005)	Question 1	**TBI group/s:** Mild to moderate (N=29) **Control group/s:** Mild to moderate "other" injuries (N= 33) and non-injured children (N= 34) **Age range of sample at recruitment:** 3-6 years	**Design:** Cross-sectional design **Time points assessed:** One time point (initial assessment once stable)	**Measure/s:** PSI (depression subscale)	**Analysis:** MANCOVA **Finding 1:** No significant differences in depression between groups (p = .215)	
Hobart-Porter et al. (2015)	Question 2	**TBI group/s:** Mild to severe (N= 125) **Control group/s:** None **Age range of sample at recruitment:** 12-17 years	**Design:** Cross-sectional design **Time points assessed**: One time point (on average 107 days after injury)	**Measure:** CESD		**Analysis**: T-test **Finding 1:** Parents of children with severe TBI have significantly higher depression scores than parents of children with mild/moderate TBI (p < .05)
Micklewright et al. (2012)	Question 1	**TBI group/s:** Moderate and severe (N=21)	**Design:** Cross-sectional design	**Measure/s:** BSI-GSI	**Analysis:** T-test **Finding 1:** Significantly more distress in TBI	

		Control group/s: OI (N= 23) **Age range of sample at recruitment:** 8-17 years	**Time points assessed:** One time point (12-36 months post injury)		groups (moderate and severe) than OI group (p < .001)	
Narad et al. (2016)	Question 1 and 2	**TBI group/s:** Severe and moderate (N=87) **Control group/s:** OI N=119: **Age range of sample at recruitment:** 3-6 years	**Design:** Prospective/ Concurrent Cohort design **Time points assessed:** 0–3, 6, 12, and 18 months after injury, and long-term follow-up an average of 6.7 years after injury	**Measure/s:** BSI-GSI and BSI-DEP. SCL -90-R, used at follow up	**Analyses:** Logistic regressions **Finding 1:** Significantly more clinically elevated distress in severe TBI group (p = .01) throughout study period **Finding 2:** No significant differences in depression between groups (p = .07), throughout study period	**Analyses:** Logistic regressions **Finding 1:** Injury severity impacted the likelihood of parents reporting clinically elevated levels of distress (p = .01) **Finding 2:** Injury severity impacted the likelihood of parents reporting clinically elevated levels of depression (p = .02)
Raj et al. (2013)	Question 2	**TBI group/s:** Mild complicated/moderate and severe group (N=117) **Control group/s:** None **Age range of sample at recruitment:** 12-17 years	**Design:** Cross-sectional design **Time points assessed:** One time point **(**1-7 months post injury)	**Measures:** CESD, SCL-90-R (general severity index)		**Analyses:** T-tests **Finding 1:** Parents of adolescents in the severe TBI group reported significantly more distress (p < .05) compared to the complicated mild/moderate group **Finding 2:** Parents of adolescents in the severe TBI group reported significantly more depression (p < .05), compared to the complicated mild/moderate group

Rivara et al. (1996)	Question 2	**TBI group/s:** Mild, moderate and severe (N=81) **Control group/s:** None **Age range of sample at recruitment:** 6-15 years	**Design:** Prospective cohort design **Time points assessed:** Baseline (asked to rate pre-injury), 3 months, 1 year, and 3 years post-injury	**Measure/s:** HIS (mental health index and depression subscale)		**Analyses:** Stepwise Regressions **Finding 1:** Mental health at 3 years predicted by pre-injury family roles ($p < .001$) **Finding 2:** Mental health at 3 years predicted by pre-injury depression ($p < .007$) **Finding 3:** Change in mental health from baseline to 3 years predicted by pre-injury rated general wellbeing ($p < .001$)
Ryan et al. (2016)	Question 1	**TBI group/s:** Mild, moderate and severe (N=78) **Control group/s:** Typically developing children (N= 40) **Age range of sample at recruitment:** 5-15 years	**Design:** Cross-sectional design **Time points assessed:** One time point (24 months post-injury)	**Measure/s:** GHQ	**Analysis:** ANOVA **Finding 1**: No significant differences in mental health between groups ($p = .096$)	
Stancin et al. (1998)	Question 1	**TBI group/s:** Moderate and severe TBI with OI (N=28) **Control group/s:** OI (N=80) **Age range of sample at recruitment:** 6-12 years	**Design:** Cross-sectional design **Time points assessed:** One time point (1 month post-injury)	**Measure/s:** BSI-GSI	**Analysis:** ANCOVA **Finding 1:** Significantly more distress in severe TBI group (compared to OI group; $p = 0.01$) **Analysis:** Multiple regression	

					Finding 2: Belonging to TBI group predicted distress ($p < .01$)	
Stancin et al. (2008)	Question 1 and 2	**TBI group/s:** Severe, moderate and complicated mild (N=89) **Control group/s:** OI (N= 119) **Age range of sample at recruitment:** 3-6 years	**Design:** Cross-sectional design **assessed:** One time point (from injury to up to 3 months post-injury)	**Measure/s:** BSI-GSI and BSI-DEP	**Analyses:** T-tests **Finding 1:** Significantly more distress in severe TBI group, compared to OI group ($p < .05$) **Finding 2:** Significantly more depression in severe TBI group, compared to OI group ($p< .05$) **Analysis:** Hierarchal regression **Finding 3**: Belonging to severe TBI group, compared to OI group, predicted distress ($p < .01$)	**Analysis:** Hierarchal regression **Finding** 1: Having an older child predicted distress in severe TBI group ($p< .05$)
Stancin et al. (2010)	Question 1	**TBI group/s:** Severe, moderate, and mild (N=99) **Control group/s:** OI (N=117) **Age range of sample at recruitment:** 3-6 years	**Design:** Concurrent cohort prospective design **Time points assessed:** Shortly after injury then 6, 12, 18 month follow-ups	**Measure/s:** BSI-GSI and BSI- DEP.	**Analysis:** T-test's **Finding 1**: Significantly more distress in severe TBI group (compared to OI group; $p = .04$). **Finding 2:** No significant differences in depression between groups ($p > .05$) **Analysis:** General linear mixed model analyses **Finding 3:** Differences between severe TBI and OI groups when social resources are low ($p = .01$), but not when resources are high ($p > .05$).	

Taylor et al. (2001)	Question 1 and 2	**TBI group/s:** Moderate and severe (N=92) **Control group/s:** OI (N= 55) **Age range of sample at recruitment:** 6-12 years	**Design:** Concurrent cohort prospective design **Time points assessed:** 3 weeks post-injury, then follow up at 6 months and 12 months	**Measure/s:** BSI-GSI	**Analyses:** ANOVA's **Finding 1:** No significant differences in distress between groups at 6 months (p = .06) **Finding 2:** Significantly more distress in moderate and severe TBI groups (compared to OI group) at 12 months (p <.05).	**Analyses:** Path analyses **Finding 1:** Severe TBI predicted more distress at 6 months (p < .01) **Finding 2:** Pre-injury rated behaviour predicted more distress at 6 months (p < .01) **Finding 3:** Distress at 6 months predicted distress at 12 months (p < .01) **Finding 4:** Behaviour problems at 6 months (predicted by severe TBI and pre-injury behaviour) predicted more distress at 12 months (p < .05)
Wade et al. (1996)	Question 1 and 2	**TBI group/s:** Moderate and severe (N=96) **Control group/s:** OI (N=69) **Age range of sample at recruitment:** 6-12 years	**Design:** Cross-sectional design **Time points assessed:** One time point (within first month of injury)	**Measure/s:** BSI-GSI, BSI-DEP and BSI-ANX.	**Analyses:** ANCOVAs **Finding 1:** Significantly more distress in TBI groups, compared to OI group (p = .01) **Finding 2:** Significantly more anxiety in TBI groups, compared to OI group (p = .01) **Finding 3:** No significant differences between TBI and OI groups in depression (p > .05)	**Analyses:** Hierarchal multiple regressions **Finding 1:** Severity of other injuries (not related to brain injury severity score) predicted psychological distress (p < .05) **Finding 2:** Maladaptive coping predicted psychological distress (p < .001)

Wade et al. (1998)	Question 1	**TBI group/s: S**evere and moderate (N=109) **Control group/s:** OI (N=80). **Age range of sample at recruitment:** 6-12 years	**Design:** Prospective design **Time points assessed:** Baseline (soon after injury), 6 months and 12 months	**Measure/s:** BSI-GSI	**Analysis:** MANCOVA **Finding 1:** Significantly more distress in severe TBI group, based on average score across time points (compared to OI group; $p < .05$) **Analyses:** Logistic regressions **Finding 2:** Higher rate of clinically severe distress symptoms in severe TBI group, compared to OI group at 6 months ($p < .05$) **Finding 3:** Higher rate of clinically severe distress symptoms in severe TBI group, compared to OI group at 12 months ($p < .05$)	
Wade et al. (2001)	Question 1 and 2	**TBI group/s:** Moderate and severe (N=103) **Control group/s:** OI (N= 71) **Age range of sample at recruitment:** 6-12 years	**Design:** Prospective design **Time points assessed:** 6 months and 12 months after baseline	**Measure/s:** BSI-GSI	**Analyses:** Hierarchical regressions **Finding 1:** Injury group (TBI vs OI) did not predict distress at 6 months ($p > .05$) **Finding 2:** Injury group (TBI VS OI) did not predict change in distress from 6 months to 12 months ($p > .05$)	**Analyses:** Hierarchical regressions **Finding 1:** Active coping at 6 months predicted distress in TBI group ($p < .01$) **Finding 2:** Humour is associated with less distress at 12 months in TBI group ($p < .01$)

Wade et al. (2002)	Question 1	**TBI group/s:** Moderate and severe (N=109) **Control group/s:** OI (N=80) **Age range of sample at recruitment**: 6-12 years	**Design:** Prospective design **Time points assessed:** Baseline, 6 months, 12 months and extended follow up (average 4.10 years later).	**Measure/s:** BSI-GSI	**Analysis:** General linear mixed model analysis **Finding 1:** No significant differences in distress between groups ($p > .05$)	
Wade et al. (2005)	Question 1	**TBI group/s:** Severe and moderate (N=100) **Control group/s:** OI (N=68) **Age range of sample at recruitment:** 6-12 years	**Design:** Prospective design **Time points assessed:** Multiple follow-ups spanning 6 years.	**Measure/s:** BSI-GSI	**Analyses:** General linear mixed model analysis **Finding 1:** No main effect of group (TBI vs OI) on distress ($p = .44$) **Analyses:** T-test's **Finding 2:** Significantly more distress in moderate TBI group compared to OI group ($p = .013$) **Finding 3:** No significant differences in distress between **s**evere TBI group and OI group ($p = .257$)	
Wade et al. (2010)	Question 2	**TBI group/s:** Moderate and severe (N= 48) **Control group/s:** OI (N=89) **Age range of sample at recruitment:** 3-6 years	**Design:** Prospective concurrent cohort design **Time points assessed:** Shortly after injury, 6, 12, and 18 months follow up	**Measure/s:** BSI-GSI		**Analysis:** General linear mixed model analyses **Finding 1:** Parent sex interacted with injury group to predict distress ($p = .003$). Fathers of children with moderate and severe TBI reported significantly higher levels of distress than mothers ($p < .05$).

Yeates, Taylor, Woodrome, et al. (2002)	Question 1	**TBI group/s:** Moderate and severe (N=97) **Control group/s:** OI (N= 55) **Age range of sample at recruitment:** 6-12 years	**Design:** Prospective design **Time points assessed:** When medically stable (baseline), 6 months, 12 months	**Measure/s:** BSI-GSI	**Analysis:** MANCOVA **Finding 1:** No significant differences in distress between groups (p = .07)	
Youngblut & Brooten (2006)	Question 2	**TBI group/s:** Mild TBI (head trauma; N=134) **Control group/s:** None **Age range of sample at recruitment:** 3-6 years	**Design:** Prospective design **Time points assessed:** Baseline (24-48 hours after hospitalisation) and 2 weeks post-discharge	**Measure/s:** MHI (Psychological distress scale)		**Analyses:** Hierarchal multiple regressions **Finding 1:** Mothers distress is predicted by social support (p < .05) **Finding 2:** Mothers distress is predicted by pre-injury psychological wellbeing (p < .05)
Youngblut & Brooten (2008)	Question 2	**TBI group/s:** Mild TBI (head trauma; N=80) **Control group/s:** None **Age range of sample at recruitment:** 3-6 years	**Design:** Prospective design **Time points assessed:** Baseline (24-48 hours after hospitalisation) and 3 months post-discharge	**Measure/s:** MHI (Psychological distress scale)		**Analysis:** Multiple linear regression analysis **Finding 1:** Psychological distress at 3 months is predicted by pre-injury psychological distress (p< .01)

Note. Exact p values are given where papers have stated them, where a p value is not provided it is represented as $p > .05$ or $p < .05$. Designs are reported as relevant to the data extracted and presented in the paper. TBI= Traumatic Brain Injury; OI= Orthopaedic Injury; ANOVA= Analysis of variance; ANCOVA= Analysis of covariance; MANCOVA= Multivariate analysis of covariance; BSI-ANX= Brief Symptom Inventory-Anxiety Symptom Scale; BSI-DEP= Brief Symptom Inventory-Depression Symptom Scale; BSI-GSI= Brief Symptom Inventory-Global Severity Index; GHQ= General Health Questionnaire; HIS=Health Insurance Survey; MHI= Mental Health Inventory; PSI= Parenting Stress Index; SCL-90-R= The Symptom Checklist-90-R

Participants. There were a mix of injury severities included in the studies; 12 studies reviewed recruited a sample of parents of children with moderate and severe TBI (Durber et al., 2017; Micklewright, King, O'Toole, Henrich, & Floyd, 2012; Narad et al., 2016; Stancin et al., 1998; Taylor et al., 2001; Wade et al., 1996, 1998; Wade et al., 2001; Wade et al., 2002; Wade et al., 2005; Wade et al., 2010; Yeates, Taylor, Woodrome et al., 2002), nine studies included parents of children with different injury severities (Anderson et al., 2013; Durish et al., 2017; Goldstrohm & Arffa, 2005; Hobart-Porter et al., 2015; Raj et al., 2013; Rivara et al., 1996; Ryan et al., 2016; Stancin et al., 2008, 2010) and three studies included parents of children with mild TBI only (Ganesalingam et al., 2008; Youngblut, & Brooten, 2006, 2008). The majority of studies that utilised control groups (in relation to review question one) recruited parents of children with orthopaedic injuries. The remaining three studies recruited typically developing children (Anderson et al., 2013; Goldstrohm & Arffa, 2005; Ryan et al., 2016), including one study that also included children with "other injuries" as a control group (Goldstrohm & Arffa, 2005).

The majority of studies recruited "primary caregivers" of children and reported that this consisted of a sample that largely included mothers. Aside from this, one study only recruited mothers (Youngblut & Brooten, 2006) and another chose mothers as an informant for two-parent families (Stancin et al., 2010). Four studies allowed for recruitment of both mothers and fathers of the same child (Goldstrohm & Arffa, 2005; Narad et al., 2016; Wade et al., 2010; Youngblut & Brooten, 2006) and in four studies it was not clear which parent/guardian took part (Anderson et al., 2013; Durish et al., 2017; Ganesalingam et al., 2008; Ryan et al., 2016).

Sample sizes varied from 21 (Micklewright et al., 2012) to 181 (Ganesalingam et al., 2008) in the TBI group. The age ranges of children were from three years (Durber et al., 2017; Durish et al., 2017; Goldstrohm & Arffa, 2005; Narad, et al., 2016; Stancin et

al., 2008, 2010; Wade et al., 2010; Youngblut & Brooten, 2006, 2008) to 17 years (Micklewright et al., 2012). The majority of studies were conducted in the US (N=22), and two studies recruited participants from Australia (Anderson et al., 2013; Ryan et al., 2016).

Design. Fifteen studies used prospective designs that used at least two time points (Durber et al., 2017; Durish et al., 2017; Ganesalingam et al., 2008; Narad et al., 2016; Rivara et al., 1996; Stancin et al., 2010; Taylor et al., 2001; Wade et al., 1996, 1998; Wade et al., 2002; Wade et al., 2005; Wade et al., 2010; Yeates, Taylor, Woodrome, et al., 2002; Youngblut & Brooten, 2006, 2008) and nine studies reported cross-sectional designs (albeit may have originally formed part of different types of designs) and reported on one time-point in their paper (Anderson et al., 2013; Goldstrohn & Arffa, 2005; Hobart-Porter et al., 2015; Micklewright et al., 2012; Raj et al., 2013; Ryan et al., 2015; Stancin et al., 1998; Stancin et al., 2008; Wade et al., 1996).

Measures. The majority of the studies used the Brief Symptom Inventory (BSI) to measure distress. Four studies used the General Health Questionnaire (GHQ; Anderson et al., 2013; Hobart-Porter et al., 2015; Raj et al., 2013; Ryan et al., 2016), one study used the depression scale of the Parenting Stress Index (PSI; Goldstrohm & Arffa, 2005), one study used the mental health index of the Health Insurance Survey (Rivara et al., 1996) and two studies used the Psychological Distress subscale of the Mental health Index (Youngblut & Brooten, 2006, 2008).

Main Findings

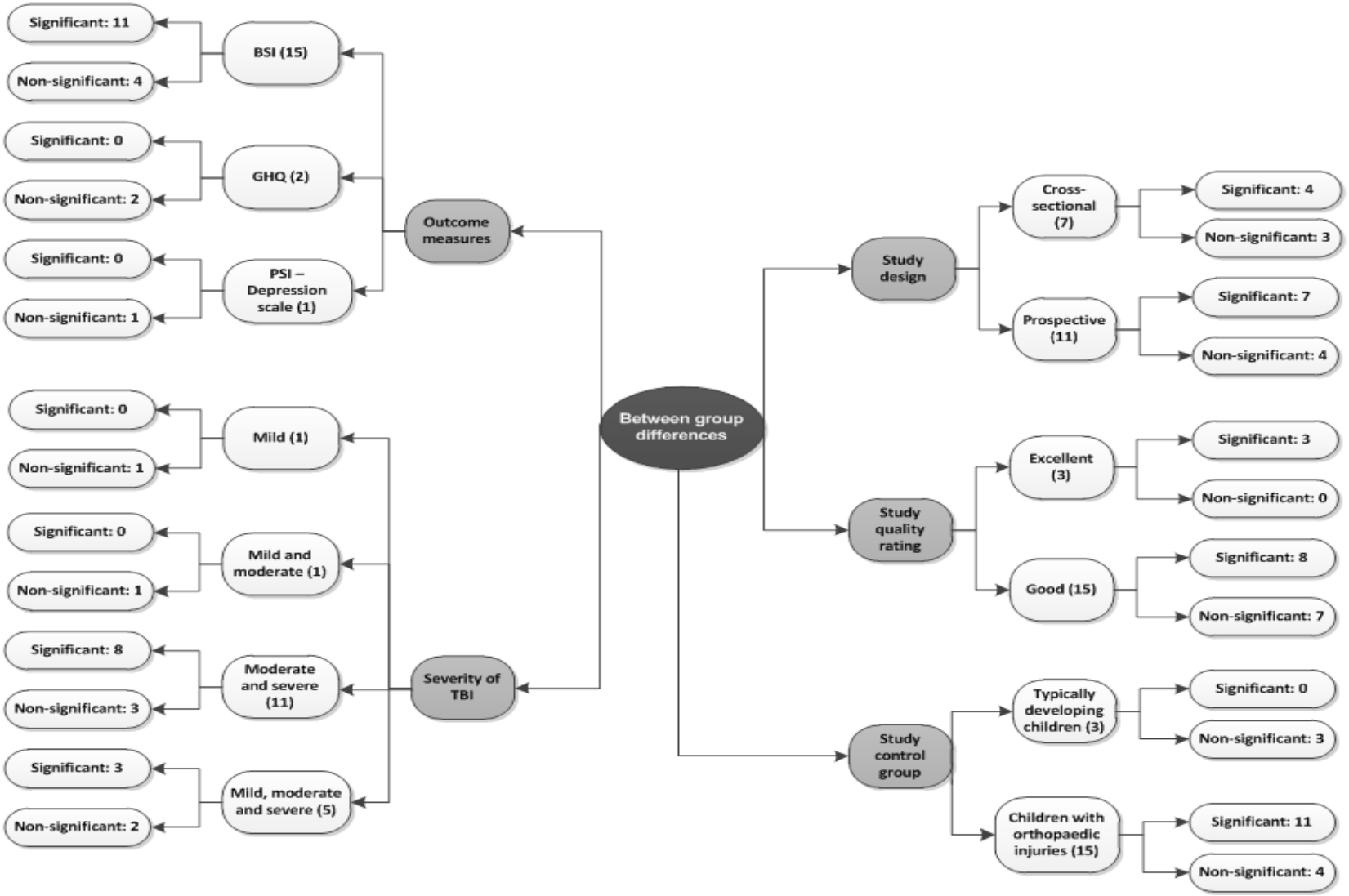
Do parents of children with TBI experience elevated levels of psychological distress compared to parents of other children?

Description of findings. Eighteen studies in the review answered this question and quality scores ranged from good to excellent. There were mixed findings; 11 studies

reported significant differences between TBI groups and controls in at least one of their analyses (Durber et al., 2017; Durish et al., 2017; Micklewright et al., 2012; Narad et al., 2016; Stancin et al., 1998; Stancin et al., 2008, 2010; Taylor et al., 2001; Wade et al., 1996, 1998; Wade et al., 2005) and seven studies reported no significant differences in all of their analyses (Anderson et al., 2013; Ganesalingam et al., 2008; Goldstram & Arffa, 2005; Ryan et al., 2016; Wade et al., 2001; Wade et al., 2002; Yeates, Taylor, Woodrome, et al., 2002).

Relationships within studies. One study assessed moderators when comparing distress between parents of children with TBI and parents of children without TBI (Stancin et al., 2010); in this study parents of children with TBI were significantly more distressed when social resources were low. Within the 11 studies that reported significant differences, five of these studies also used an analysis that found no differences between groups. Three of these studies did not find differences when measuring depression, but did when measuring general distress (Narad et al., 2016; Stancin et al., 2010; Wade et al., 1996). One study did not find differences at the six-month analysis but did at the 12-month analysis (Taylor et al., 2001). Finally, one study did not find differences between the severe TBI group and control group but did between the moderate TBI group and control group (Wade et al., 2005).

Between-group similarities and differences. Due to the heterogeneity in the studies reviewed, studies were grouped based on major participant/study characteristics, and whether they had at least one significant finding when comparing parents of children with TBI to parents of other children, or no significant findings (see Figure 2).



F*igure 2.* Between-group differences for studies comparing parents of children with TBI to other parents. BSI=Brief Symptom Inventory. GHQ=General Health Questionnaire. PSI=Parenting Stress Index.

Severity of TBI. Eight out of the 11 studies that found significant differences between groups only recruited parents of children with moderate or severe TBI (Durber et al., 2017; Durish et al., 2017; Micklewright et al., 2012; Narad et al., 2016; Stancin et al., 1998; Taylor et al., 2001; Wade et al., 1996, 1998; Wade et al., 2005) and, therefore, the majority of the significant findings relate to those with moderate or severe TBI. Furthermore, six of the 11 studies that reported significant differences concluded that the differences in distress were regarding the severe TBI group and control group, rather than the mild or moderate groups (Durber et al., 2017; Durish et al., 2017; Narad et al., 2016; Stancin et al., 1998; Stancin et al., 2008, 2010).

Outcome measures. All studies included in the review that found significant differences between parents' distress reported the use of the BSI; studies that used other outcome measures did not report significant differences (Anderson et al., 2013; Goldstrohn & Arffa, 2005; Ryan et al., 2016). The majority of studies that reported significant differences between parents used the global distress index of the BSI. However, one study that utilised the anxiety index and one study that utilised the depression index found significant differences between parents (Stancin et al., 2008; Wade et al., 1996).

Study design. A mix of significant and non-significant results were found across studies utilising both cross-sectional and prospective designs. Only one prospective study found significant results at one time-point (12 months) but not the other time point assessed (6 months; Taylor et al., 2001).

Study control group. Three studies did not recruit parents of children with orthopaedic injuries as a control group (Anderson et al., 2013; Goldstrohm & Arffa, 2005; Ryan et al., 2016) and included a sample of typically developing children instead; none of these studies found significant differences between groups.

Study quality rating. There were not substantial differences in the quality ratings of studies that found significant differences to studies that didn't, and all studies received good quality ratings overall. However, studies that received a quality rating of excellent were more likely to report significant differences between groups.

Which factors significantly impact the psychological distress of parents of children with TBI?

Description of findings. Eleven of the studies included in the review reported significant factors that impact on parental distress (Hobart-Porter et al., 2015; Narad et al., 2016; Raj et al., 2013; Rivara et al., 1996; Stancin et al., 2008; Taylor et al., 2001; Wade et al., 1996; Wade et al., 2001; Wade et al., 2010; Youngblut & Brooten, 2006, 2008) and there were 21 significant results amongst these studies. Quality ratings for this review question ranged from fair to excellent.

Relationships within studies. Five of the 11 studies that reported significant findings also reported on non-significant findings, using the same type of statistical analysis (Stancin et al., 2008; Wade et al., 1996; Wade et al., 2001; Youngblut & Brooten, 2006, 2008). These non-significant findings related to both parents' characteristics (such as parental coping, support, stressors, concerns; Stancin et al., 2008; Wade et al., 2001; Youngblut & Brooten, 2006, 2008), children's characteristics (such as head injury severity and sickness of child; Wade et al., 1996; Youngblut & Brooten, 2006, 2008) and demographic factors (such as sociodemographic status, number of children and parents in the household; Wade et al., 1996; Youngblut & Brooten, 2006, 2008).

Between group similarities and differences. In examining the 21 significant factors that impact parental distress, 10 were related to children's characteristics and 11 were related to parent/family factors (see Figure 3). Quality ratings differed slightly;

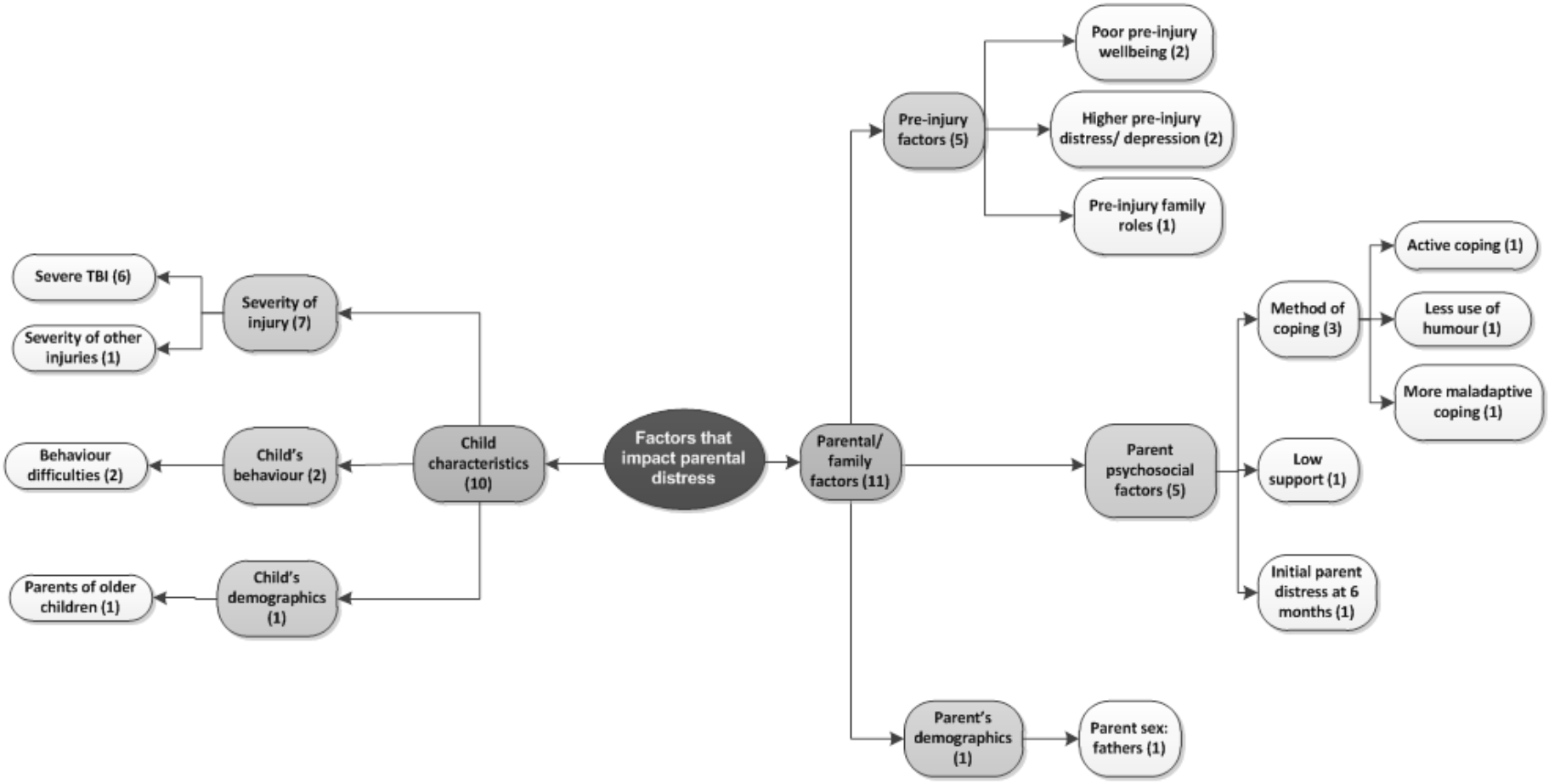


Figure 3. A visual representation of factors that increase parental distress.

studies that reported on children's characteristics ranged from good to excellent and studies that reported on parental/family variables ranged from fair to good.

Children's characteristics. As seen in Figure 3, six analyses found that there was more distress in parents of children with severe TBI (Hobart-Porter et al., 2015; Narad et al., 2016; Raj et al., 2013; Taylor et al., 2001) and one analysis (Wade et al., 1996) found that the severity of other injuries significantly predicted more distress. Two analyses found that children's behaviour impacted on parental distress. One analysis found pre-injury behaviour contributed to increased parental distress (Taylor et al., 2001) and another analysis found that post-injury behaviour contributed to increased parental distress (Taylor et al., 2001). In addition, one analysis found that parents of older children were more likely to be distressed (Stancin et al., 2008).

Parental/family factors. As per Figure 3, there were 11 significant analyses related to parental/family variables that impact distress. Five analyses found that pre-injury factors were shown to increase distress, such as pre-injury wellbeing, pre-injury distress and pre-injury family roles (Rivara et al., 1996; Youngblut & Brooten, 2006, 2008). However, it is of note that two of these studies received a quality rating of fair (Rivara et al., 1996; Youngblut & Brooten, 2006) and, therefore, these findings should be reviewed in the context of this. Five analyses found that current parent circumstances, related to psychosocial factors, were important; coping was found to be a significant factor in predicting distress in three analyses (Wade et al., 1996; Wade et al., 2001), lower social support predicted mother's distress in one study (Youngblut & Brooten, 2006), and distress at six months was predictive of distress at 12 months in another study (Taylor et al., 2001). Finally, one study found that parental sex impacted distress; in this study fathers of children with moderate and severe TBI experienced more distress than mothers (Wade et al., 2010).

Discussion

The narrative review set out to examine psychological distress in parents of children with TBI and intended to explore factors that significantly impact on parental distress. Overall it appears that parents of children with TBI are at risk of experiencing psychological distress, with more than half of the studies reviewed reporting significantly more distress in parents of children with TBI, compared to control groups. The review supports a range of factors that could impact parental distress. The findings will be considered below and the results will be reviewed in relation to literature in the area.

More than half of the studies reviewed (61%) reported at least one analysis that demonstrated significantly more distress in parents of children with TBI, compared to parents of other children. These findings align with the literature, with research reporting that parents of children with TBI experience poorer family functioning (Anderson et al., 2013; Rashid et al., 2014) and significant injury-related burden (Wade et al., 1998). These findings are also consistent with qualitative research that has highlighted the detrimental impact that paediatric TBI can have on parents' psychological health (Brown et al., 2013; Du Toit et al., 2013).

Significant differences in distress between parents of children with TBI and parents of children without TBI were more likely to be reported when using the general distress index of the BSI. The BSI general distress index includes a range of presentations of distress, such as somatization and hostility. It is possible that this measure was able to capture a broader range of emotional reactions that were pertinent to the parents in the studies reviewed. Indeed, qualitative studies have documented a range of emotions, such as anger and loss (Brown et al., 2013) and these emotions may not be fully captured in other scales. Studies which included samples of those with severe injuries were more likely to find significant differences between parents; this is consistent with other research

which has found that families of those with severe TBI are more negatively impacted (Rashid et al., 2014). All studies that recruited typically developing children as a control group, rather than children with orthopaedic injuries, did not find significant differences. It is of note that all studies that recruited parents of typically developing children compared their distress levels to parents of children with mild TBI. According to this review parents of children with mild TBI may be less distressed than parents of children with moderate to severe injuries; this might account for the non-significant findings when comparing them to parents of typically developing children.

In the review it was evident that not all studies found that parents of children with TBI were more distressed; this is in keeping with a review by Rashid et al. (2014), in which it was reported that not all families report family dysfunction. Other research has reported that there are no differences in parenting stress (Hawley et al., 2003; McKinlay, Albicini, & Than, 2018) or family functioning (Anderson et al., 2001; Ryan et al., 2016) between parents of children with TBI and other parents. The differences in distress levels between parents could partly be related to individual risk and resistance factors, as outlined by Wallander and colleagues' model (Wallander et al.,1989; Wallander & Varni, 1998). This model highlights various factors that might increase vulnerability to poor mental health and may account for the variability in the psychological health of parents of children with TBI in this review.

The second review question examined which factors significantly impact on parental distress. The studies reviewed highlight the importance of children's characteristics, such as their severity of injury (Hobart-Porter et al., 2015) and behaviour (Taylor et al., 2001). This is consistent with research that has found that neurobehavioral problems and level of participation can predict caregiver emotional distress (Sander et al., 2013) and quality of life (Koskinen, 1998). There were also characteristics related to

parents and families that appeared to impact on parental distress, such as pre-injury wellbeing (Youngblut & Brooten, 2006, 2008) and current coping mechanisms (Wade et al., 2001). Similar findings have also been reported in assessing caregivers of adults with TBI, with studies reporting that pre-morbid psychiatric difficulties and current psychological factors predict caregivers' adjustment and distress (Sander et al., 2013; Verhaeghe, Defloor, & Grypdonck, 2005).

As noted above, there were a variety of factors associated with parental distress in the studies reviewed; some of these factors appeared to be consistent with the model of caregiver adjustment proposed by Wallander et al. (1989) and Wallander and Varni (1998), described above. However, it was evident that there was not one unanimous set of factors that impacted parental distress. Indeed, several of the studies reviewed conducted analyses that found that some characteristics of parents, children and families, such as the child's severity of injury (Youngblut & Brooten, 2006) and parental social support (Youngblut & Brooten, 2008), did not significantly impact parental distress. The differences between study findings might be partly explained by the differences in sample characteristics. In addition, there is significant heterogeneity in recovery in childhood TBI (Narad et al., 2017) and this may contribute to what is most distressing for parents at a particular time.

Critique of Studies Reviewed

There were many strengths in the studies reviewed, such as the use of prospective designs and robust statistical analyses. None of the studies reviewed received a quality rating of poor, reducing bias in the studies reviewed. However, the quality analysis also indicated some important limitations. The most common weaknesses were not addressing how missing data was dealt with, not controlling for potential confounding variables (such as demographic variables) and not reporting exact significance values. A number of these

issues could have increased bias in the studies reviewed. Indeed, it has been reported that not identifying or controlling for confounder variables and having missing data can lead to bias (Kang, 2013; Pannucci & Wilkins, 2010).

It is also noteworthy that the diversity of the participants in the samples appears to have been somewhat limited. The majority of the studies reviewed included mothers in their sample of primary caregivers; these studies may, therefore, of overlooked fathers' needs. In addition, all but two of the studies recruited parents in the US. TBI care can vary significantly between countries; for example, the way that care is provided and paid for can differ significantly (Cnossen et al., 2016). It is possible that differences between the care provided between countries has a significant impact on families and, therefore, it would be beneficial to have studies that include samples across the globe.

Clinical Implications

The findings from the review are tentative but suggest that parents of children with TBI may be more likely to experience distress. Parents' distress may put them at risk of future mental health problems and could have a detrimental impact on their children (Narad et al., 2017). Family factors have been shown to impact TBI trajectories (Narad et al., 2017) and studies have demonstrated that parental mental health and behaviour can predict social and behavioural outcomes of children with brain injury (Catroppa et al., 2017; Treble-Barna et al., 2016). Due to the potential impact of parental distress, it would be beneficial for parents' mental health to be routinely assessed in clinical services and, where necessary, be signposted to or offered psychological support. In addition, due to the bi-directional relationships between parents and children (Taylor et al., 2001), family interventions may be appropriate. Family interventions have shown to be successful in improving parental distress after paediatric brain injury (Braga, Da Paz Junior, & Ylvisaker, 2005; Wade, Carey, & Wolfe, 2006). In understanding parents' adjustment

clinicians may also want to consider the factors that increase parents' risk of distress and provide family-focused support as necessary.

Future Research

Many of the studies reviewed included mothers and it would be valuable to include both caregivers, so that both parents needs can be assessed. It would also be useful to conduct more research outside of the US to understand parents' experiences within different healthcare systems. Some studies were excluded from the review because they included the control samples within their analysis of factors that impact distress. Consequently, future studies would benefit from obtaining a larger sample so that control groups and TBI groups can be assessed separately. In addition, due to the changing and complex nature of family adjustment after TBI it would be beneficial for studies to continue to undertake longitudinal research. The review also indicated that there may be differences in distress between parents of children with mild TBI, compared to parents of children with severe TBI. It would be beneficial for future research to be more homogenous and it is possible that a larger focus should be on parents of children with moderate to severe injuries.

Strengths and Weaknesses of the Review

The current review is the first review, to the authors' knowledge, to specifically focus on parents' psychological distress following paediatric TBI; therefore, the review has made a unique addition to the evidence base and provided an up-to-date critique of studies in this area. The review also examined which factors may impact distress, as well as examining parents' distress in comparison to other parents; this has provided a broader understanding of parental mental health after paediatric TBI. However, there are still limitations that need to be acknowledged. One limitation is that the studies included in the review were heterogeneous in the samples they included, whilst this helped to provide an

overall review of the research area, it also may have impacted the variability of results. Another limitation is that the review found no evidence of unpublished studies that met inclusion criteria and, therefore, the review could have included a publication bias.

Similar to the review on family functioning by Rashid et al. (2014), a number of the studies included in this review were not independent of each other and used overlapping cohorts, as indicated above. None of these studies reported the exact same statistic and, therefore, no analysis was repeated in the review. Nevertheless, this limits the overall representativeness and generalisability of the review and indicates the need for more independent studies.

Conclusions

Overall, the findings of this narrative review suggest that parents of children with TBI may experience significant emotional reactions, particularly parents of children with severe TBI. However, not all parents experience elevated distress and there are a wide range of factors that might increase parents' risk of distress, such as parents' coping style, pre-injury mental health and factors related to the child's current presentation (Taylor et al., 2001; Wade et al., 2001; Youngblut & Brooten, 2006, 2008). The review demonstrates the complex nature of parental adjustment to TBI and the individuality of parents; this underlines a need to take a family-centred approach to understanding families' adjustment and risk. Future research assessing distress in parents of children with TBI is warranted.

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Chapter 2. Bridging Chapter

Results from the Systematic Review

As reported in the systematic review, a large number of parents of children with Traumatic Brain Injury (TBI) experience significantly higher levels of distress compared to parents of other children; this is particularly true for parents of children with severe TBI. The review indicated that there were a number of factors that significantly impacted parental distress. These were related to child characteristics, such as behaviour (Taylor et al., 2001) and age of child (Stancin et al., 2008), and parent characteristics or family resources, such as type of coping (Wade et al., 1996; Wade et al., 2001) and social support (Youngblut & Brooten, 2006). The findings of the systematic review were in keeping with research that has demonstrated the significant impact of brain injury on families. Indeed, research has found that paediatric TBI can significantly increase family burden (Wade et al., 1998) and can be detrimental to family functioning (Anderson et al., 2013; Rashid et al., 2014).

The systematic review has added to the wider literature around parents' mental health following paediatric illness. Research regarding parental distress in relation to child illness is rapidly expanding as researchers acknowledge that medical events happen to children who are located within families (Kazak et al., 2005). Pinquart (2017) recently conducted a meta-analysis and reported that parents of ill children are significantly more stressed. Furthermore, research has found that parents of ill children may be at an increased risk for poor mental health (Davidson, Jones, & Bienvenu, 2012; Needle, O'Riordan, & Smith, 2009; Shudy et al., 2006; Woolf, Muscara, Anderson, & McCarthy, 2016) and life threatening illnesses in children can lead to parental anxiety, depression and acute traumatic stress (Manne et al., 2004; Muscara et al., 2015; Needle et al., 2009; Nelson & Gold, 2012).

Distress in Parents of Children with Cancer

The systematic review demonstrated the impact that a particular paediatric injury, TBI, may have on parents' mental health, and examined which factors impact this. Another area that has recently started to expand within paediatric health is the impact of paediatric cancer on parents' mental health. A paediatric cancer diagnosis can have a long-term impact on parents (Yalug et al., 2011) and it is unsurprising that parents can experience extreme stress in the context of this (Woodgate, Tailor, Yanofsky, & Vanan, 2016). Parents of these children have reported high levels of anxiety, depression and post-traumatic stress disorder (PTSD; Bruce, 2006; Harper et al., 2014; Norberg & Boman, 2008; Vrijmoet-Wiersma et al., 2008).

Within the oncology literature, research has investigated post-traumatic stress responses in parents following children's diagnosis and/or treatment (Brown, Maden-Swain, & Lambert, 2003; Bruce et al., 2011). The 4th edition of the DSM broadened the understanding of PTSD and stipulated that it can be triggered by being told one's child has a life-threatening condition (American Psychiatric Association, 1994). A recent study found four factors related to post-traumatic stress in parents of children with cancer: dysphoria, re-experiencing, hyperarousal and avoidance (Cernvall, Alaie, & von Essen, 2011). However, the use of the diagnostic term "PTSD" in parents of children with cancer is contentious (Kangas et al., 2002), due to the complex experiences of being a parent of a child with cancer. A cancer diagnosis involves possible future threats, such as re-occurrence of the cancer (Cernvall et al., 2011) and Bruce (2005) suggests that parents may never reach a "post" position. Consequently, instead of referring to the diagnosis of PTSD, others have used the term "post-traumatic stress symptoms" (PTSS; Bruce et al., 2011; Kangas et al., 2002).

Two reviews have been conducted in relation to PTSS in parents of children with cancer. These reviews have noted a range of different factors that can increase parental risk of PTSS (Bruce, 2006; Yalug et al., 2011). In a review by Bruce (2006) a range of factors that impact parental PTSS were summarised, such as parental sex (Haegen & Luminet, 2015), social support (Brown et al., 2003; Kazak, Barakat, Meeske, & Christakis, 1997), family functioning (Brown et al., 2003) and parent-child concordance in symptoms (Barakat et al., 1997). A more recent review conducted by Yalug et al. (2011) reported a range of similar factors to Bruce (2006), such as type of coping (Greening & Stoppelbein, 2007), social support (Greening & Stoppelbein, 2007), cognitive avoidance (Norberg, Pöder, & von Essen, 2011), pre-morbid psychiatric diagnosis (Yalug et al., 2008), time since diagnosis (Jurbergs, Long, Ticona, & Phipps, 2007) and child prognosis (Yalug et al., 2008). Both of these reviews concluded that children's diagnoses and treatment can have a long-term impact on parents and parents' PTSS may be impacted by a number of factors.

Empirical Study

In reviewing research in the area of paediatric health, it is evident that more research needs to examine the mental health needs of parents of children with illnesses or injuries. In particular, parents of children with cancer are at risk of poor mental health (Harper et al., 2014) and can experience post-traumatic stress (Bruce et al., 2011); consequently, it would be beneficial to conduct a study assessing PTSS in these parents. Two reviews have assessed post-traumatic stress reactions in parents of children with cancer and acknowledged that studies in this area are too heterogenous and have recruited parents of children with a range of different cancer diagnoses (Bruce, 2006; Yalug et al., 2011). Kazak et al. (2005) recommends that further research is needed to identify sub-groups of families and risk factors to help develop suitable interventions. To address this

problem, the empirical paper will focus on parents of children with brain tumours; only two studies have explored PTSS in this population (Bruce et al., 2011; Fuemmeler, Elkin, & Mullins, 2001) and, therefore, the research in this area still needs developing. In assessing parents' PTSS, it is important to consider both mothers and fathers; this is a weakness of studies examining parents of children with cancer (Kazak, 2005; Yalug et al., 2011). To this end, the empirical paper will address the current gaps in the literature and explore PTSS in mothers and fathers of children with a brain tumour.

Chapter 3. Empirical paper

Prepared for submission to 'Children's Health Care'*

*****Author guidelines can be seen in Appendix D. It is of note that margins and spelling requirements are in line with UEA formatting guidance but will be altered when submitting the paper. In addition, DOI numbers have been removed from the references, in line with journal guidance.

Post-traumatic stress in parents of children with a brain tumour

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Post-traumatic stress in parents of children with a brain tumour

Research has documented that parents of children with cancer can experience psychological difficulties following their child's diagnosis and treatment, including experiencing post-traumatic stress symptoms. This study set out to assess post-traumatic stress in parents of children with a brain tumour and sought to examine factors that impact post-traumatic stress. The study found high levels of post-traumatic stress in parents of children with a brain tumour and found that disengaged coping was related to post-traumatic stress symptoms. Clinicians should be aware of post-traumatic stress reactions in these parents and more research is needed to evaluate interventions in this population.

Introduction

Approximately 102,000 people in the UK are living with a brain tumour (The Brain Tumour Charity, n.d.). Brain tumours are the leading cause of cancer-related death in those under 40 (Brain Tumour Research, 2016) and one of the most common causes of cancer-related death in children (Cancer Research UK, n.d.). Medical advances mean that survival rates for brain tumours have increased (Moore, 2005), however children with a brain tumour can go on to have long-term disabilities (Lannering, Marky, Lundberg, & Olsson, 1990). The impact of the brain tumour itself, or the treatment, can lead to "late effects" and cognitive impairments (Srivastava, Pandey, & Meena, 2016; The Brain Tumour Charity, n.d; Woodgate, Tailor, Yanofsky, & Vanan, 2016). Children who have had a brain tumour have been reported to have poorer physical health (Zebrack et al., 2004), poorer neurocognitive sequelae (Moore, 2005; Mulhern, Merchant, Gajjar, Reddick, & Kun, 2004) and greater behavioural and emotional difficulties (Upton & Eiser, 2006) than other children.

Considering the reports above, it is not surprising that research has documented that the impact of a childhood brain tumour is a negative and stressful experience for both parents and children (Bennett, English, Rennoldson, & Starza-Smith, 2013; Shortman et al., 2013; Woodgate et al., 2016). Parents of children with brain tumours can experience considerable uncertainty (Woodgate et al., 2016) and fear for the future (Shortman et al., 2013). A range of emotional outcomes have been reported for parents of children with cancer, such as anxiety, depression and post-traumatic stress (Bruce, 2006; Norberg & Boman, 2008; Pinquart, 2018; Soanes, Hargrave, Smith, & Gibson, 2009; Vrijmoet-Wiersma et al., 2008).

Post-traumatic stress in parents of children with cancer

Post-traumatic stress disorder (PTSD) is a particular mental health concern among parents of children with cancer (Kazak, Alderfer, Rourke et al., 2004; Ljungman et al., 2014; Smith, Redd, Peyser, & Vogl, 1999) and researchers have reported on post-traumatic stress presentations in these parents. For example, Norberg, Lindblad, and Boman (2005) reported that intrusive thoughts and images associated with their child's diagnosis was a commonly reported symptom in parents of children who had received cancer treatment. Prevalence rates of PTSD in parents of children with cancer range from 6-25% (Bruce, 2006), compared to figures of 4% in the general adult population (NHS Digital, 2014).

Following the research reported above, PTSD models have been utilised to explore trauma reactions of parents in paediatric settings (Bakker, Van der Heijden, Van Son, & Van Loey, 2013; Horsch, McManus, & Kennedy, 2012). For example, Ehlers and Clark's (2000) cognitive model has been used to explore trauma in families of children with cancer (D'Urso, Mastroyannopoulou, Kirby, & Meiser-Stedman, 2018; Kangas, Henry, & Bryant, 2002). This model highlights the nature of trauma memories in increasing current threat, as well as individual appraisals about the trauma and/or its sequalae. The model suggests that the memory of the trauma, and the subsequent appraisals, can be impacted by several factors, such as cognitive processing during the trauma, characteristics of the trauma and coping. Additionally, attempts to control threat symptoms can prevent change in the memory and appraisals of the trauma.

Whilst Ehlers and Clark's (2000) cognitive model has a good evidence base (see Brewin & Holmes, 2003), it was not developed in a medical setting. It has been reported that parents' experiences of having a child diagnosed with cancer is unique (Bruce, 2006) and parents of children with cancer may never reach a "post" position, due to ongoing

threat (Bruce, 2005). Consequently, it is important to explore research conceptualising trauma in this specific population. In referring to cancer-related trauma, researchers have used the term post-traumatic stress symptoms (PTSS; Bruce, Gumley, Isham, Fearon, & Phipps, 2011). The pediatric medical traumatic stress model was specifically developed to examine medical traumatic stress for families (Kazak et al., 2005). This model suggests that families are impacted by the immediate experiences of the event, and then experience on-going demands and challenges, such as those related to the treatment. It underlines the importance of subjective experiences, over medical factors, in shaping psychological outcomes. The model continues to be supported by studies within paediatric health (Price, Kassam-Adams, Alderfer, Christofferson, & Kazak, 2015) and has been applied in understanding post-traumatic stress reactions of parents of children with cancer (Dunn et al., 2012; Patiño-Fernández et al., 2008).

Risk factors for cancer-related PTSS

As indicated above, there are potential long-term implications of brain tumours on children (Moore, 2005; Mulhern et al., 2004) and researchers have reported that parents of these children may be at risk of developing PTSS (Bruce et al., 2011; Fuemmeler, Mullins, & Marx, 2001). Consequently, it is important to explore PTSS in this population further and to investigate which factors increase parents' risk of developing PTSS. Reviews assessing research into PTSS in parents of children with cancer note a range of different factors that can increase PTSS (Bruce, 2006; Yalug, Tufan, Doksat, & Yaluğ, 2011). These reviews, and subsequent research, have noted a relationship between PTSS and demographic and medical factors, such as parents' pre-morbid psychiatric diagnoses (Yalug et al., 2008), parental sex (Haegen & Luminet, 2015), child's age (Kazak et al., 1998), days in hospital (Landolt, Ystrom, Sennhauser, Gnehm, & Vollrath, 2012) and time since treatment (Jurbergs, Long, Ticona, & Phipps, 2007). However, others report mixed

findings for these factors and suggest they may be less important than other factors (Bruce, 2006; Kazak et al., 2005).

The pediatric medical traumatic stress model (Kazak et al., 2005) highlights the importance of the subjective experiences of parents and underlines the value of targeting psychosocial factors, such as social support. A range of psychosocial variables have shown to correlate with or predict PTSS, such as type of coping (Greening & Stoppelbein, 2007), family functioning (Brown, Madan-Swain, & Lambert, 2003), self-efficacy (Best, Streisand, Catania, & Kazak, 2001) and social support (Brown et al., 2003). Social support is a psychosocial factor which has not been explored in parents of children with a brain tumour. However, research reports that it can contribute to the development and maintenance of PTSD (Brewin, Andrews, & Valentine, 2000; Cordova, Riba, & Spiegel, 2017; Guay, Billette, & Marchand, 2006) and has shown to be related to PTSS in parents of children with cancer (Brown et al., 2003; Kazak et al., 1998). Social support can change cognitive appraisals (Joseph, Williams, & Yule, 1997) which may then influence a parent's sense of threat, thereby impacting PTSD as explained in the model by Ehlers and Clark (2000).

Bruce (2006) reports that the impact of coping strategies in parents of children with cancer has been under-explored and, more recently, research has suggested that method of coping can impact PTSS in parents of children with cancer (Greenberg & Stoppelbein, 2007; Tremolada, Bonichini, Schiavo, & Pillon, 2012). To date, the impact of coping styles has been explored in parents of children with brain tumours (Bruce et al., 2011; Fuemmeler et al., 2001), however disengaged coping in relation to the child's illness and treatment has not been investigated in these parents. Disengaged coping can include avoidance, denial and wishful thinking (Compas, Champion, & Reeslund, 2005). Disengagement strategies, such as suppression, can result in greater intrusions (Wegner,

Schneider, Knutson, & McMahon, 1991), which is related to greater risk of PTSD symptoms (Ehlers & Clark, 2000).

In keeping with the model of medical traumatic stress, and the prevention of PTSS, on-going cancer-related issues are important to explore (Kazak, 2005, 2006; Kazak et al., 2005). In regard to the impact of brain tumours specifically, it has been reported that children show more behavioural difficulties (Upton & Eiser, 2006). Furthermore, studies have reported associations between parental PTSS and emotional and behavioural difficulties in children (Bruce, 2005; Davis, Parra, & Phipps, 2010; Nakajima-Yamaguchi et al., 2016). It is possible that children's behaviour may increase parents' sense of threat and, using the cognitive model (Ehlers & Clark, 2000), this could increase parents' risk of developing PTSS.

Current study

As seen above, a plethora of research has been conducted to investigate PTSS in parents of children with cancer and reviews report that between 6-44% of parents have high levels of PTSS (Bruce, 2006; Ljungman et al., 2014; Yalug et al., 2011). However, research in this field has been criticised for using populations that are too heterogenous (Bruce, 2006; Bruce et al., 2011; Yalug et al., 2011). Only two studies have examined factors that impact PTSS in parents of children with a brain tumour (Bruce et al., 2011; Fuemmeler et al., 2001); these studies found that parent-child conflict, tumour re-occurrence and illness uncertainty increased the risk of PTSS (Bruce et al., 2011; Fuemmeler et al., 2001). These studies were limited in the number of variables they could explore and, consequently, there are still important variables that remain unexplored. In addition, these studies only included parents of children off treatment and it is important to explore PTSS in parents of children on treatment too.

First, this study will explore the proportion of parents of children with a brain tumour who experience high levels of PTSS. The study will then explore the relationship between PTSS and demographic and medical factors. Research on the importance of these factors appears to be mixed, as indicated above, and therefore it would be beneficial to explore the impact of these factors further in parents of children with a brain tumour. The study will also explore the impact of three psychosocial variables on PTSS; these variables will focus on factors that have not been fully explored in this population and will build on the work of Bruce et al. (2011) and Fuemmeler et al. (2001) by exploring social support, children's behaviour and disengaged coping. These variables are fitting with the literature cited above, as well as models of PTSS (Kazak, 2006; Kazak et al., 2005) and PTSD (Ehlers & Clark, 2000). Finally, this study will conduct a supplementary analysis to explore differences in PTSS between parents of children with a brain tumour (using PTSS scores in this study) and parents of children with other types of cancer and paediatric injuries (using PTSS scores reported in two published studies; Norberg & Boman, 2013; Nugent, Ostrowski, Christopher, & Delahanty, 2006).

Research questions:

1) What proportion of parents of children with a brain tumour experience PTSS?
2) Are there significant differences in parental PTSS depending on medical/demographic variables?
3) Does social support, disengaged coping and children's behaviour impact on parents' PTSS?

Supplementary research question:

4) Do parents of children with a brain tumour show significantly different levels of PTSS compared to parents of children with other medical conditions (as reported in other studies)?

Materials and method

Design

A within-groups cross-sectional quantitative design was used to examine factors that impact PTSS.

Participants

Participants were parents/guardians of children (aged 4-16) receiving care from a paediatric oncology service, who had been diagnosed with a brain tumour at least six months ago and had not completed treatment more than five years ago (if they had not received treatment, they were not diagnosed more than five years ago). Parents of children who were terminally ill and parents who were unable to speak English were not asked to participate.

Procedure

Study recruitment took place between June 2018-January 2019. The study was advertised through a tertiary care NHS hospital and online. Initially, parents at the hospital site who met the study criteria were sent a letter informing them that the study was taking place; this letter was attached with an information sheet and parents could contact the research team at this stage. Parents were also approached about the study at a tertiary hospital, or one smaller local hospital, and were initially approached by a member of the clinical team. Parents had the option of completing the study via paper questionnaires, or online, and could do the study either in clinic or at home. The online

version of the study was advertised throughout the recruitment period and advertised through Facebook, Twitter, one webpage and two charity mailing lists.

Parents of 126 children at the tertiary care hospital site were sent a letter informing them of the study and 34 eligible participants (of 25 children) were recruited; therefore, the response rate for recruitment at the hospital site was 20%. Twenty-four parents (of 24 children) were recruited through online recruitment. Overall, a total of 58 parents (41 mothers, 14 fathers and three whom did not specify their relationship), of 49 children completed the study.

Measures

Demographic information

Basic demographic information about the child and the parents were collected. Parent details included: age, ethnicity, occupation, relationship to child, previous PTSS and psychological therapy. Details about the child included: age, sex, ethnicity, type of brain tumour, date diagnosed, stage of treatment, date of last treatment, type of treatment, frequency of brain scans and re-occurrence of the brain tumour.

Coping Strategies Inventory Short Form

The Coping Strategies Inventory (CSI; Tobin, Holroyd, & Reynolds, 1984) includes statements about different types of coping and asks individuals how much they have used these strategies on a five-point scale (from none to very much) when experiencing a particular event. Parents were asked to rate their coping in respect to their child's brain tumour diagnosis and/or treatment. The CSI is reported to be a valid and reliable measure, with internal reliability ratings ranging from .71-.94 (Tobin et al., 1984; Tobin, Holroyd, Reynolds, & Wigal, 1989). The scale has been used in parents of children with cancer previously (Trask et al., 2003). This study used the validated short-

form version (Addison et al., 2007) and used the disengagement sub-scale used in previous research (Mendonca, 2010; Speyer et al., 2016), which includes coping responses related to wishful thinking, denial, avoidance and self-blame. The measure has internal reliability ratings of between .58 -.72 for the subscales being used in this study (Mendonca, 2010).

Multidimensional Scale of Perceived Social Support

The Multidimensional Scale of Perceived Social Support (Zimet, Dahlem, Zimet, & Farley, 1988) contains 12 statements about the current support individuals receive and asks parents to rate how much they agree with each statement on a seven-point scale (from strongly disagree to strongly agree). The scale can be divided into support from family, friends and significant others and provides an overall score. The measure yields a Cronbach's alpha of .93 and is shown to be valid and reliable measure (Canty-Mitchell & Zimit, 2000; Zimet, Powell, Farley, Werkman, & Berkoff, 1990) and has been used with parents of children with cancer previously (Bayat, Erdem, & Gül Kuzucu, 2008).

The Child and Adolescent Behavior Inventory

The Child and Adolescent Behavior Inventory (CABI) is a measure that examines different domains of psychopathology in children and young people (Burns, Taylor, & Rusby, 2001). The study used the oppositional defiant disorder subscale (Burns, Lee, Servera, McBurnett, & Becker, 2015); this has eight questions related to children's' behaviour and is rated on a six-point scale (from almost never to almost always). The measure was chosen as it is free to use and has been used and validated in research assessing children from four to 18 years old (Burns, Moura, Beauchainem, & McBurnett, 2014; Lee, Burns, & Becker, 2017). It has good validity (Burns et al., 2008; Moura, 2001) and reliability (Gomez, Burns, Walsh, & Hafetz, 2005), with an internal consistently rating of .88 (Gomez et al., 2005).

Impact of Events Scale-Revised

The Impact of Events Scale-Revised (IES-R; Weiss & Marmar, 1997) was used to examine parents' PTSS in relation to their child's diagnosis and/or treatment. It lists difficulties parents might have experienced in the last seven days, in respect to a specific event; in the current study parents were asked to answer these questions in relation to their child's brain tumour diagnosis and/or treatment. It asks parents how much they have been bothered by these difficulties on a five-point scale (from not at all to extremely). The scale contains 22 questions, including three sub-scales (intrusion, avoidance, and hyperarousal) and yields an overall score. This measure has shown to have good reliability and validity (Creamer, Bell, & Failla, 2003) and has been used in a population of parents with children with cancer (Bruce et al., 2011; Davis et al., 2010). It has been reported to have internal reliability ratings of .95 for the total score (Davis et al., 2010) and ratings of .94, .87 and .91 for the intrusion, avoidance and hyperarousal sub-scales respectively (Creamer et al., 2003).

Ethical issues

The research was conducted following the British Psychological Society (BPS; 2010) guidelines for the conduct of psychological research. The project was approved by the Social Care Research Ethics Committee and obtained Health Research Authority approval and Research and Development approval. Participants were asked to provide consent and were reminded of their right to withdraw from the study. Participants confidentiality was maintained in line with the Data Protection Act (1998) and The General Data Protection Regulation (GDPR; 2016).

Analyses

The questions were analysed on Statistical Package for Social Sciences (SPSS; version 23) and the data was screened for errors and missing values. Assumption testing

was conducted in line with relevant analyses. To analyse the impact of potential confounding variables, differences between those recruited online, compared to those recruited from the NHS site, were analysed using an independent samples *t*-test.

To analyse the relationship between demographic variables and PTSS, seven independent sample *t*-tests, one Mann-Whitney *U* test and two Spearman's correlations were performed. To examine the impact of psychosocial variables on PTSS, multiple regression analyses were conducted using the enter method. Four multiple regressions were conducted in total (one regression for total PTSS total score and then a regression relating to each sub-scale: hyperarousal, intrusion and avoidance). Finally, the PTSS scores of parents in this current study were compared to the PTSS scores (using the IES-R) reported in a study which included parents of children with other forms of cancer (Norberg & Boman, 2013) and a study which included parents of children with paediatric injury (Nugent et al., 2006) using a one-sample *t*-test. To control for multiple testing the Holm-Bonferroni method (Holm, 1979) was used for research questions two and four.

Results

Preliminary analyses

A total of 58 parents (of 49 children) took part in this study. For research questions two to four, only one parent of each child was included in the analysis (N=49), due to the requirement of independence in observations used in these analyses. A small amount of questionnaire data was missing (0.16%) and Little's (1988) Missing Completely at Random test (MCAR) was not significant (x^2 = 165.1, *df* = 164, *p*= .461). Subsequently, mean imputation was used for the missing data. Method of recruitment was explored as a potential confounding factor (online recruitment vs NHS recruitment), but had no significant impact on parental PTSS (*t*(41.03) = -1.36, *p* = .19). Assumption tests

were carried out and in instances where assumptions were not met data was transformed or non-parametric analyses were conducted.

Sample characteristics

The main characteristics of the sample can be seen in Table 3 and Table 4. Thirty-six percent of parents identified as having received psychological therapy in relation to their child's diagnosis and/or treatment and 31% identified as experiencing a prior traumatic event, before their child's cancer diagnosis, that resulted in PTSS symptoms. A summary of the means and standard deviations for the main outcome measures are presented in Table 5.

Table 3

Demographic characteristics of parents

Characteristics of parents (N=58)	N (%)	Mean (*SD*)
Relationship to child		
Mother	41(70.67)	
Father	14 (24.13)	
Other[a]	3 (5.17)	
Age		41.84 (7.12)
Ethnicity		
White British	50 (86.21)	
Other[b]	8 (13.79)	
Occupation[c]		
Managers	11 (18.97)	
Professionals	19 (32.76)	
Technicians and associate professionals	1 (1.72)	
Clerical support workers	3 (5.17)	
Service and sales workers	4 (6.90)	
Craft and related trade workers	4 (6.90)	
Plant and machine operators and assemblers	3 (5.17)	
Elementary occupations	1 (1.72)	
Other (e.g. retried, full-time carer, homemaker)	12 (20.69)	

[a]Refers to parents who did not specify or identified themselves as a 'guardian'. [b]Refers to parents who described themselves as the following: White British/Irish, Scottish, Indian, Black British African, White British African, Welsh and did not specify. [c]Occupation was categorised in accordance with the international standard classification of occupations (International Labour Organization, 2010).

Table 4

Demographic characteristics of children

Characteristics of children (N=49)	N (%)	Mean (*SD*)
Age		9.55 (3.81)
Sex		
Female	26 (53.06)	
Male	23 (46.94)	
Ethnicity		
White British	44 (89.80)	
Other[a]	5 (10.2)	
Diagnosis		
Medulloblastoma	12 (24.49)	
Astrocytoma	12 (24.49)	
Craniopharyngioma	7 (14.29)	
Glioma	5 (10.20)	
Ependymoma	4 (8.16)	
Other	9 (18.37)	
Treatment type		
Chemotherapy	9 (18.37)	
Surgery	6 (12.24)	
Proton therapy	2 (4.08)	
Mix of more than one treatment	25 (51.02)	
No treatment indicated/did not specify	7 (14.29)	
Time since diagnosis (months)		41.57 (25.6)
Time since treatment (months)		20.03 (18.67)
Stage of treatment		
Long-term follow up	34 (69.39)	
On treatment	11 (22.45)	
Watching/waiting for treatment	4 (8.16)	
Frequency of brain scans		-
6 months or more	28 (57.14)	
Less than 6 months	19 (38.78)	
Did not specify	2 (4.08)	

[a]Refers to parents who indicated one of the following: Indian, mixed race, Welsh, White British/Irish and did not specify.

Table 5

Scores for the main outcome measures

Outcome measure scores[a]	Mean	*SD*
IES-R total score	42.51	20.90
CABI (oppositional defiance disorder subscale)	9.84	9.16
Multidimensional scale of perceived social support	56.51	15.17
CSI (disengaged coping subscale)	24.96	7.24

[a]These scores relate to parents who were included in the main analyses

Main results

What proportion of parents of children with a brain tumour experience PTSS?

Table 6 shows parents scores on the IES-R. Overall, 76% of parents scored as having high levels of PTSS symptoms (>24) and 60% of parents scored above the "cut-off" for PTSD (>33; as set by Creamer et al., 2003). The intrusion subscale had the highest overall mean score.

Table 6

IES-R scores for the whole sample

	IES-R Avoidance mean (mean, SD)	IES-R Intrusion mean (mean, SD)	IES-R Hyperarousal mean (mean, SD)	IES-R Total score (mean, SD)	Parents with high PTSS scores (%)	Parents scoring above the cut-off for PTSD
Mothers (N=41)	1.81 (0.92)	2.29 (0.98)	1.88 (1.12)	44.51 (19.65)	88%	68%
Fathers (N=14)	1.18 (0.90)	1.19 (0.92)	0.68 (0.94)	23.93 (18.95)	36%	29%
Total sample (N=58)	1.71 (0.94)	2.01 (1.06)	1.58 (1.17)	39.84 (21.12)	76%	60%

Are there significant differences in parental PTSS depending on medical/demographic variables?

To analyse the impact of medical and demographic variables, seven independent-samples *t*-tests, one Mann-Whitney *U* and two Spearman's correlations were conducted, following relevant assumptions testing. As indicated in Table 7, only one analysis was significant (parent age) and this became non-significant when the Holm-Bonferroni method was applied for multiple comparisons. Estimates of the effect sizes were all small (see Table 7).

Table 7

Relationship between PTSS and medical/demographic variables

	Test statistic	Significance (*p* value)	Holm-Bonferroni correction	Effect size	Effect size description
Time since diagnosis[a]	$t = .18$	.862	-	$d = 0.05$	Small
Time since treatment	$r_s = .03$	.873	-	$r = 0.03$	Small
On/off treatment	$t = 1.35$	.184	-	$d = 0.48$	Small
Type of treatment[b]	$t = -.73$	.468	-	$d = 0.28$	Small
Number of re-occurrences[c]	$t = -.75$	.455	-	$d = 0.25$	Small
Frequency of brain scans[d]	$t = .30$	.767	-	$d = 0.09$	Small
Child age[e]	$t = .35$	.725	-	$d = 0.10$	Small
Child sex	$t = -.14$	.891	-	$d = 0.04$	Small
Parent age	$r_s = -.29$	.047	.005	$r = 0.29$	Small
Mothers/fathers	$U = 96.5$	.227	-	$r = 0.29$[f]	Small

[a]Less than 3 years since diagnosis compared to more than 3 years. [b]One type of treatment compared to more than one type of treatment. [c]No re-occurrences compared to one re-occurrence. [d]Scans less than every 6 months compared to scans every 6 months and more. [e]Children aged 4-10 compared to children aged 11-16. [f]Effect size calculated from z-score

Does social support, disengaged coping and children's behaviour impact on parents' PTSS?

Following assumption testing, four multiple regression analyses were carried out to understand the impact of social support, disengaged coping and children's behaviour on PTSS. The regression models were significant for: the IES-R total score ($F(3, 45) = 5.84$, $p = .002$), showing a medium effect size and explaining 23% of the variance in PTSS; the IES-R intrusion score ($F(3, 45) = 3.66$, $p = .019$), showing a medium effect size and explaining 14% of the variance in intrusion symptoms; the IES-R avoidance score ($F(3, 45) = 6.93$, $p = .001$), showing a large effect size and explaining 27% of the variance in avoidance symptoms; the IES-R hyperarousal score ($F(3, 43) = .38$, $p = .015$), showing a medium effect size and explaining 15% of the variance in hyperarousal symptoms. In assessing the significance of the independent variables separately, disengaged coping was

the only variable that added significantly to the regression models in the analyses for total PTSS, avoidance and hyperarousal symptoms. Disengaged coping and social support added significantly to the model exploring intrusion symptoms. The regression coefficients and standard errors can be seen in Table 8 and regression models can be seen in Table 9.

Table 8

Multiple regression analyses: Regression coefficients and standard errors

	Disengaged coping[b]			Social support[c]			Behaviour[d]		
	B	SE_b	β	*B*	SE_b	β	*B*	SE_b	β
IES-R total score[a]	1.39	.37	.50***	.26	.18	.19	.39	.28	.18
IES-R Intrusion	.43	.16	.39**	.15	.07	.29*	.12	.12	.14
IES-R Avoidance	.59	.14	.55***	.06	.07	.11	.11	.11	.13
IES-R Hyperarousal	.39	.13	.42**	.06	.06	.14	.13	.10	.18

[a]Higher scores on the IES-R indicate higher PTSS. [b]Higher scores on disengaged coping measure indicate higher use of disengaged coping strategies. [c]Higher scores on social support measure indicate higher levels of perceived social support. [d]Higher scores on the behaviour measure indicate more behavioural difficulties.
*p < .05. **p < .01. ***p < .001

Table 9

Multiple regression analyses: overall regression model

	R^2	Adjusted R^2
IES-R total score	.28**	.23
IES-R intrusion	.20*	.14
IES-R avoidance	.31**	.27
IES-R hyperarousal	.21*	.15

*p <.05. **p <.01.

Do parents of children with a brain tumour show significantly different levels of PTSS compared to parents of children with other medical conditions (as reported in other studies)?

Scores from the IES-R used in the main analyses (M = 42.51, SD = 20.09) were compared to the IES-R scores from a study assessing parents of children with cancer (Norberg & Boman, 2013; M = 17.8, SD = 13.81) and a study assessing parents of children with paediatric injury (Nugent et al., 2006; M = 12.2; SD = 15.50). A one sample t-test showed the IES-R score in parents of children with a brain tumour was statistically higher than parents of children with cancer by a mean of 18.94, $t(48) = 8.61$, $p < .001$ and was statistically higher than parents of children with paediatric injury by a mean of 24.71, $t(48) = -10.56$, $p < .001$. The Holm-Bonferroni method was applied for multiple comparisons ($p < .0025$ and $p < .05$); following this both tests were still significant. Estimated large effect sizes were found for both comparisons (d = 1.23, d = 1.51, respectively).

Discussion

Overall the study found a high proportion of PTSS in parents of children with a brain tumour, with 76% of parents showing high levels of PTSS. The study set out to assess factors that impacted on PTSS. A range of variables were studied, including demographic variables, medical variables and psychosocial variables. The findings showed that disengaged coping was the only factor significantly related to overall PTSS. The study used a supplementary analysis to compare differences in the levels of PTSS between parents included in this study and a study which included parents of children with all types of cancer (Norberg & Boman, 2013) and a study which included parents of children with paediatric injury (Nugent et al., 2006). Results indicate that parents of children with a brain tumour may be at a higher risk of PTSS than parents of children with

other types of cancer, or paediatric injuries. The results of the study will first be summarised in the context of existing literature and then the clinical implications, future research directions and limitations of the study will be discussed.

The study found that 76% of parents reported high levels of PTSS and 60% scored above the cut-off for PTSD. In the previous studies that have assessed PTSS in parents of children with brain tumour, it was reported that 29% (Bruce et al., 2011) and 43% (Fuemmeler et al., 2001) met criteria indicative of PTSD. The high prevalence of PTSS found in this study supports previous research that suggests this population may be a particularly vulnerable sub-group (Bruce, 2006). It is possible that this study found higher levels of PTSS than the previous studies, above, due to including parents of children still in treatment.

The results show that none of the demographic or medical variables investigated were significantly related to PTSS. The evidence for the impact of these factors appears to be mixed, according to reviews in this area (Bruce, 2006; Yalug et al., 2011), with some researchers suggesting that objective factors may be less important than subjective factors (Kazak et al., 2005). It is of note, however, that Bruce et al. (2011) found that tumour re-occurrence was related to PTSS in parents of children with brain tumour. Additionally, in this study the results showed that younger parents had higher PTSS levels, although this was not significant when multiple comparisons were controlled for. Consequently, the impact of medical and demographic variables on PTSS in this population warrants further investigation.

The study set out to examine if social support, disengaged coping and children's behaviour affected parents' PTSS. All final models were significant and explained between 14% to 27% of the variance in PTSS. Disengaged coping (measuring denial, avoidance, wishful thinking and self-blame) was related to overall PTSS, as well as

avoidance, intrusion and hyperarousal symptoms. This finding is in keeping with previous research that has shown that avoidant coping predicts PTSD severity (McNeill & Galovski, 2015; Silver, Holman, McIntosh, Poulin, & Gil-Rivas, 2002) and is related to PTSS in parents of children with cancer (Fuemmeler et al., 2001; Greening & Stoppelbein, 2007; Norberg, Pöder, & von Essen, 2011). In the cognitive model of PTSD (Ehlers & Clark, 2000) avoidant coping strategies, such as suppression, are important in preventing change in the trauma memory and appraisal and can impact on current threat.

Social support did not significantly add to the model assessing overall PTSS, or avoidance and hyperarousal symptoms. However, it did significantly add to the model assessing intrusion symptoms, in which more social support predicted more intrusions. The finding is in contrast to previous research that has indicated that social support is related to less PTSS in parents of children with cancer (Brown et al., 2003; Kazak et al., 1998). However social support was measured differently in the above studies; therefore, it is possible that differences in the way support is measured might contribute to different results. Another hypothesis is that the helpfulness of social support can depend on the support offered (Ullman, 1999). This might help to understand the mixed findings in the research; for example, one study found that increased support from friends can increase risk of PTSD (Scarpa, Haden, & Hurley, 2006) and another study found that social support had no significant impact on PTSS in mothers of children with cancer (Pelcovitz et al., 1996).

The study found that parents' reports of children's behaviour was not related to their level of PTSS in any of the regression analyses, despite research that has reported associations between parental PTSS and behavioural difficulties in children who have had cancer (Bruce, 2005; Davis et al., 2010; Nakajima-Yamaguchi et al., 2016). It is possible that behaviour may have general effects on parenting distress and wellbeing, as noted in

previous research (Klassen et al., 2007; Patterson, Holm, & Gurney, 2004) but does not affect PTSS specifically. However, these results are tentative and require further exploration.

Finally, the study set out to tentatively compare PTSS in parents of children with a brain tumour in this study, to PTSS in parents of children with any type of cancer (Norberg & Boman, 2013) and paediatric injury (Nugent et al., 2006); the results show significantly higher levels of PTSS in parents included in this study. It has been suggested that parents of children with cancer are a "high risk" group, due to the threat to life and experience more trauma than parents of children with "low risk" conditions (Landolt, Boehler, Schwager, Schallberger, & Nuessli, 1998). Indeed, research shows that trauma severity (Brewin et al., 2000) and perception of life threat (Holbrook, Hoyt, Stein, & Sieber, 2001) can increase risk of PTSD, possibly putting parents of children with cancer diagnoses at an increased risk. Furthermore, children with brain tumours experience more late effects compared to other cancer diagnoses (Lannering et al., 1990; Srivastava et al., 2016) and a brain tumour diagnosis is associated with higher mortality (Cancer Research UK, n.d.). Together, these factors could make the experience of a brain tumour diagnosis especially traumatising.

Implications for clinical practice

Whilst the results from this study are tentative, the findings indicate that it would be beneficial to educate health care professionals about the high rates of trauma in this population. In addition, the results suggest that parents might benefit from trauma-based interventions. National Institute of Health Care and Excellence (NICE, 2018) guidelines recommend a range of treatments for PTSD, such as cognitive therapy, which have shown to be effective in reducing post-traumatic stress symptoms (Cusack et al., 2016).

However, research has not examined the efficacy of these interventions in parents of children with cancer and it is of note that parents in this setting can present with PTSS without meeting diagnostic criteria for PTSD or acute stress disorder (Kazak et al., 2005).

Kazak (2006) presents a more specific model (the pediatric psychosocial preventative health model) that is aimed at supporting families of ill children in medical settings. In this model the largest percentage of families require basic universal support and a small percentage may require mental health support. Kazak et al. (2005) has highlighted an intervention aimed at providing mental health support to children and families with cancer, the surviving cancer completely program (Kazak et al.,1999); this program incorporates cancer-related issues and uses elements of cognitive-behavioural therapy. Research has reported that this program is effective in reducing PTSS in parents of children with cancer (Kazak et al., 1999; Kazak, Alderfer, Streisand et al., 2004). In clinical practice, clinicians also need to be aware of factors that could increase parents' vulnerability to experiencing PTSS. In this study disengaged coping was shown to be a risk factor; this might indicate that parental coping is important for clinicians to identify and support parents with.

Limitations and future research directions

This study has made a unique contribution to the literature; it is the first study to examine PTSS, and factors that impact PTSS, in parents of children with a brain tumour whilst children are on or off treatment. Nevertheless, the study also has several important limitations. One of the main limitations is that the study recruited a relatively small sample and, consequently, the results are tentative. According to a priori calculations, some of the analyses conducted in this study were under-powered and may have only been able to detect large effects. However, despite the small sample size, the study recruited a similar number of participants to previous studies in this area (Bruce et al., 2011;

Fuemmeler et al., 2001) and the final sample size might reflect challenges to recruitment in this population. Future studies would benefit from inputting more time and resources into research in this area (e.g. by conducting multi-site studies and including a longer period of recruitment), in order to recruit a bigger sample.

The study assessed parents' PTSS in comparison to PTSS in parents of children with other types of cancer (Norberg & Boman, 2013) and paediatric injury (Nugent et al., 2006), reported in previous studies. The samples included in these studies did not precisely match the demographic characteristics of the sample used in the current study, therefore this is a limitation of this analysis. It was not feasible to use a matched group design in the current study; however, this type of design should be a consideration for future research in this area. In addition, the study examined the impact of children's behaviour on parental PTSS, however it is important to note that children with a brain tumour can also show cognitive and emotional difficulties (Moore, 2005; Mulhern et al., 2004; Upton & Eiser, 2006). Consequently, the impact of these difficulties may warrant further investigation. McCauley et al. (2012) provides a good overview of recommended measures for children with brain injuries that should be considered in future research.

The study set out to include both mothers and fathers of the same child in order to explore parents' experiences separately. However, the final sample size recruited meant these analyses could not be conducted and, therefore, the study largely reflected the experiences of mothers. It would be valuable for future research to focus on fathers as they are under-represented in this type of research (Kazak, 2005). In addition, the study is limited in that it used a cross-sectional design; this meant that parents' reactions over-time could not be fully explored and the study cannot draw causal conclusions. It is apparent that parents' needs may change over-time (Kazak et al., 2005) and, therefore, it is essential that longitudinal studies are carried out to track families' adjustment. Finally, considering

the high amounts of PTSS reported in this population, more research is needed to help to develop interventions aimed at these parents (Kazak, 2005).

Conclusions

Overall, the current study found high rates of PTSS in parents of children with a brain tumour. A range of factors were explored to understand which factors might increase parents' vulnerability and only disengaged coping style was found to affect overall PTSS. These results indicate that disengaged coping could be an important factor for clinicians to be aware of in this population, although the results are tentative and should be explored further. More research examining trauma in parents of children with a brain tumour is warranted, as well as further research evaluating the efficiency of interventions for these parents.

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Chapter 4. Extended Methodology

Sample Recruited

The inclusion/exclusion criteria for the study is provided in more detail below.

Inclusion criteria. The study included parents/guardians of children with any brain tumour who were receiving care from a paediatric oncology service. The study included parents of children from six months post-diagnosis to five years post-treatment completion and children could be on or off treatment. Both previous studies conducted in this area included children off active treatment (Bruce et al., 2011; Fuemmeler et al., 2001), however it was deemed to be important to include parents of children on or prior to treatment too and assess PTSS in the earlier stages post-diagnosis. Both of the studies conducted in this population used children older than eight years (Bruce et al., 2011; Fuemmeler et al., 2001), however the current study included parents of children from 4-16 years in order to understand the experiences of parents with younger children as well.

Exclusion criteria. Parents of children who were terminally ill or who were unable to speak English were not asked to participate. Parents of children who were diagnosed less than 6 months ago or were more than five years post treatment (or more than five years post-diagnosis if they had not had treatment) were excluded in order to reduce the heterogeneity in the sample. However, it is of note that one parent had a child whom was just under six months post-diagnosis. Prior to approaching this parent, it was believed that the child was six months post-diagnosis. After discussion with the research supervisor it was agreed the parent's data was still clinically useful and, therefore, they were included; this was documented as a minor protocol deviation.

Procedure

The study was advertised through one tertiary hospital and online. The recruitment procedures will be described in more detail below.

Recruitment via letters. After ethical approval was in place, instructions were provided for oncology staff at one main tertiary NHS hospital. Staff identified all potential participants and sent them a letter informing them about the study, attaching information sheets. When parents received the letter, they had the option of contacting the chief investigator (CI) about the study directly (via email) or completing a consent to share contact details form. If parents expressed an interest to take part, the CI sent a consent form with the questionnaires (either in paper form or online) and two stamped addressed envelopes (SAEs) to send back the questionnaires and consent forms separately.

Recruitment at the hospital. Parents were also recruited at the hospital by the CI or by a member of the oncology team. When the CI recruited, parents were approached by a member of the clinical team first. If parents wished to take part they were asked to provide informed consent and then were given a study pack. Parents completed the study at the hospital or took the questionnaires home and sent them back in a SAE. Parents were also offered the option of completing the study online, using a pre-generated link. If parents had consented but did not send the study questionnaires back after a month, they were sent one reminder email if they had provided consent for this. In addition, the hospital advertised the study via posters in two waiting rooms. Information sheets and consent to share contact details forms were also handed out at one local hospital, as some parents attended this hospital more frequently than the tertiary hospital.

Online recruitment. The online version of the study (conducted using Bristol Online Survey) was also posted on a webpage, Facebook pages and Twitter. The study advertisement was posted on these pages (outlining the inclusion/exclusion criteria), with the link to the online study. The study information from the advertisement was also emailed and advertised through the mailing list of two charities. The recruitment pathways utilised in this study can be seen in further detail in Figure 4.

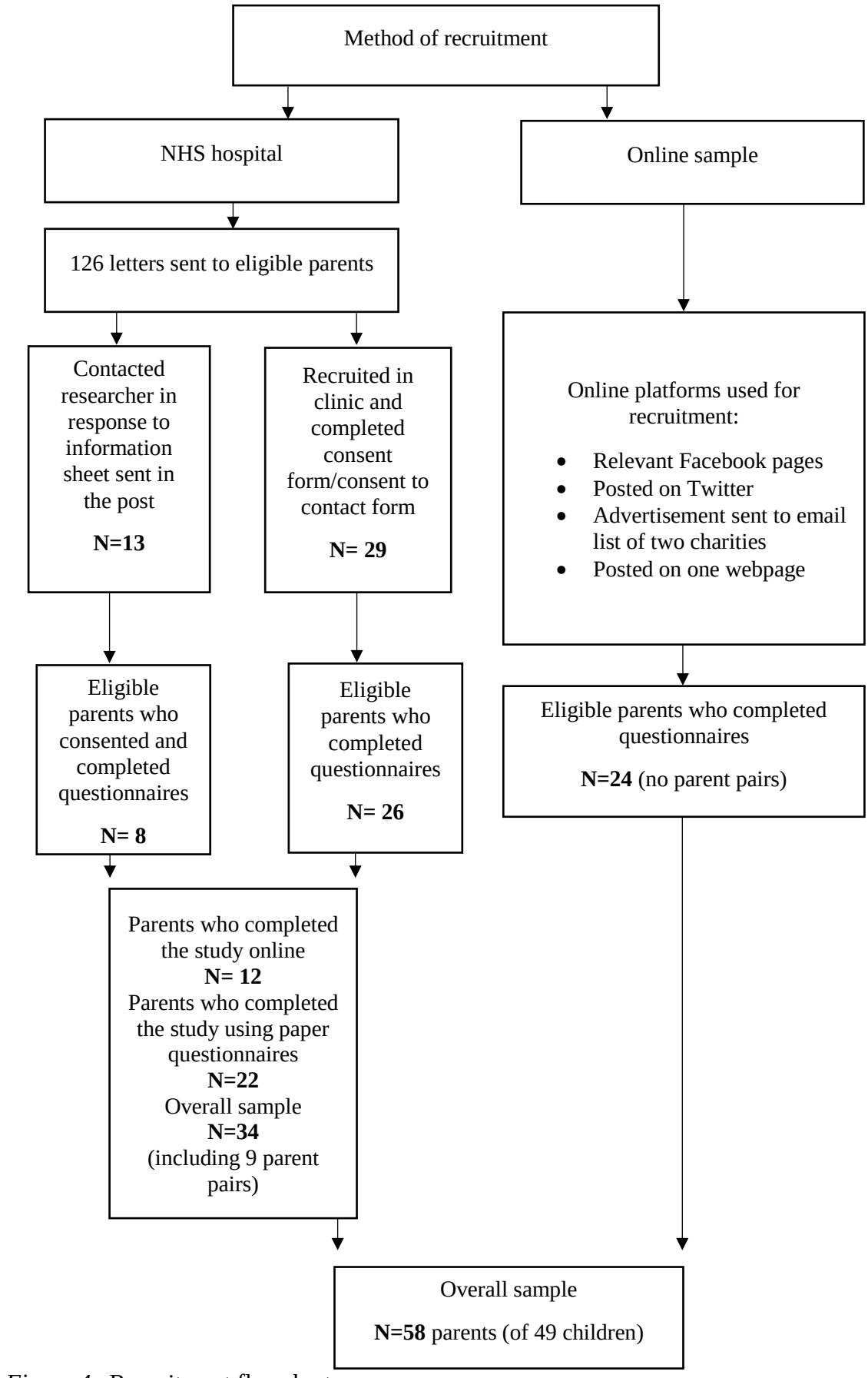

Figure 4. Recruitment flowchart

Recruitment Strategy

Based on the sample sizes obtained from the previous studies that have been conducted in this population (Bruce et al., 2011; Fuemmeler et al., 2001), the recruitment strategy for this study was considered in significant detail. It was predicted that by inviting both parents to take part, the experiences of mothers and fathers could be explored separately. However, recruiting both mothers and fathers can be a challenge (Macfadyen, Swallow, Santacroce, & Lambert, 2011), therefore the study was devised to make it more feasible for both parents to take part. This included allowing participants to do the study online or via paper questionnaires, either at home or in clinic. In addition, due to some parents not regularly attending the hospital site, letters were sent to parents. Posters were also displayed in hospital waiting rooms to advertise the study more widely and act as a potential reminder for parents. Parents who consented to be reminded about the study were sent a follow up email after a month if they had not returned the questionnaires.

In addition to NHS recruitment, online recruitment also took place in order to increase the representativeness of the study. Akard, Wray, and Gilmer (2015) recruited parents of children with cancer through Facebook advertisements, due to the challenges of recruiting families in this setting. They found that this was a cost-effective approach, and therefore, it was anticipated that online advertisement could help with recruitment in the current study.

Amendments During Recruitment

Study recruitment was monitored throughout and it was evident that the study was recruiting fewer participants than anticipated, therefore various amendments were made. One major amendment included changing the inclusion/exclusion criteria from including parents of children up to five years post-diagnosis to including parents of children up to

five years post-treatment completion. Bruce et al. (2011) found high levels of PTSS even years after treatment and, therefore, it was considered that this change would not impact on the validity of the study. Another change included handing information sheets and consent to share contact details forms out to one local hospital, where some patients attended more regularly. In addition, a change was included to allow parents to consent to the study on the same day as reading the information sheet, rather than waiting 48 hours if they had not read the information sheet; this made it practically easier for parents who did not attend the hospital regularly. A minor amendment was also made to share the study on social media/the internet more widely (such as by using Twitter) and the study received approval for two charities to send the online advertisement to their parent mailing list. Finally, the recruitment period window was extended to allow for more parents to take part.

Ethical Issues

Informed consent. The information sheets contained information about what the study would involve and provided an email address if parents had any questions. Parents were advised that they could take their time to think about the study before taking part and were then asked to provide formal consent.

Right to withdraw. Participants were reminded of their right to withdraw up until the point that the data got analysed; this information was given to participants on the information sheet. They were also reminded that they could stop the study at any time and their child's care would not be affected.

Distress. Participants were informed on the information sheet that they may get upset when completing the questionnaires and were informed that this could be a potential disadvantage of the study and reminded that they do not have to take part. They were also asked to stop completion of the questionnaires if they felt distressed. For those recruited

from the hospital, contact details of whom they could contact were given on the debrief sheet. In addition, parents recruited from the hospital sites who scored above the cut-off for PTSD on the IES-R were sent a letter advising them to see their GP and were given the contact details for clinicians at the hospital. Parents recruited via the online recruitment methods were anonymous and were provided with online resources on the information sheet and the debrief sheet to signpost them to more information and support.

Confidentiality. In line with the Data Protection Act (1998) and The General Data Protection Regulation (GDPR, 2016), participants were informed that their information would be kept confidential. Questionnaires and consent forms were held securely for the duration of the study. The questionnaires only had ID numbers and were stored separately to consent forms. There was a database with parents' ID numbers and names; this database was kept on a secure encrypted NHS pen drive for the duration of the study.

Coercion. Participants were informed that they were under no obligation to take part and that participation in the study would not impact on their child's care.

Sample Size Calculations

The target sample sizes were calculated based on the minimum sample sizes required. Ideally, the study sought to recruit more than the minimum numbers calculated, however the sample sizes of the previous research conducted were reviewed (53 and 28 respectively; Bruce et al., 2011; Fuemmeler et al., 2001) and, considering this, the study anticipated that it might only be able to recruit enough participants to detect large effects. Consequently, sample size calculations were conducted in the context of previous recruitment in this population by using large effect sizes within calculations.

It was anticipated that questions two and four would use *t*-tests and correlational analyses. Due to this study including a different population of parents to previous studies,

previous effect sizes may not be accurate and, instead, calculations using G-power 3 (Faul, Erdfelder, Lang, & Buchner, 2007) were conducted and power tables provided by Clark-Carter (2010) were utilised. To detect at least large effect sizes with .80 power and alpha at .05, it was calculated that 26 participants would be required for correlations (two-tailed), 26 participants per group would be required for *t*-tests (two-tailed) and 15 participants would be required for one sample *t*-tests (two tailed).

Question three used a multiple regression to assess the impact of three psychosocial variables on PTSS. The power of a regression decreases with the addition of variables (Clark-Carter, 2010); due to this, the study was designed to only investigate the value of three main variables. There are multiple approaches authors use for calculating sample sizes in multiple regression (Clark-Carter, 2010; Milton, 1986). For example, Green (1991) suggests recruiting 50 people, plus eight multiplied by the number of variables. Others suggest that the number of participants should be equal to the number of predictors plus 50 (Harris, 1985). Alternatively, it has been indicated that 15 participants are needed per a variable (Clark-Carter, 2010; Stevens, 2002).

As above, the study used novel research questions in an under-researched sample, therefore it was deemed that sample size calculations based on previous literature may not be entirely accurate. If conducting sample size calculations using G-power 3 (Faul et al., 2007), and tables provided by Clark-Carter (2010), to detect at least large effects sizes, with .80 power and alpha at .05, approximately 36 participants would be required. Based on the various suggestions for sample sizes noted above, and the sample size calculations using G-power 3, the study set out to recruit a minimum number of 36 participants; however, ideally, it aimed to collect up to 74 parents, using the formulae outlined by Green (1991).

Multiple Comparisons

To control for multiple testing, it is recommended that significance values are altered to help decrease type 1 errors (Abdi, 2007; Schaffer, 1995). Bonferroni corrections are widely used but are susceptible to type 2 error, increasing the chance the null hypothesis will be incorrectly accepted (Holm, 1979). Instead, adjusted Bonferroni calculations are recommended as being more powerful, such as the Holm-Bonferroni method (Bender & Lange, 2001; Wright, 1992). In this study, the Holm-Bonferroni method (Holm, 1979) was used to control for multiple testing for questions two and four.

Effect Sizes

There are debates as to whether effect sizes should be calculated for non-significant findings (Grissom & Kim, 2011), however for the purpose of this paper recommendations from Field (2009) were followed which suggest that effect size calculations are performed for non-significant analyses too. It was anticipated that supplying estimates of effect sizes, even for non-significant findings, could help guide future research in this area. Descriptors for effect sizes were followed in reference to guidelines by Cohen (1988).

Parametric Assumption Testing

Parametric assumptions were checked for all analyses below, in line with recommendations according to each statistical test (Carter-Clarke, 2010; Field, 2009; Laerd Statistics, 2015a). To check that data was normally distributed, a variety of statistical techniques were used. Visual inspections were carried out on histograms and QQ plots were screened. In addition, Sharpio-Wilk's test was used to assess normality and skewness and kurtosis values were assessed and converted into Z-scores to determine significant violations of normality. Field (2009) suggests that Z-scores above or below

1.96 indicate significant deviations from normality. Outliers were examined using box plots and any identified outliers were inspected in further detail.

Analyses and Assumption Testing

It is of note that in the analyses that will be described below, the data of only one parent of each child was included, due to the assumption of independence in observations in the analyses (except from the analysis comparing parent pairs). In circumstances in which two parents of one child took part, parents who identified themselves as the primary caregiver were chosen. For parents who both described themselves as primary caregivers, one parent was chosen completely at random.

Are there significant differences in parental PTSS depending on medical/demographic variables? To assess the significance of demographic variables, it was anticipated that independent samples *t*-tests and Pearson's correlations would be performed. Relevant assumptions testing in line with parametric testing, as described above, were carried out. In addition to this, homogeneity of variance was assessed using Levene's tests for equality of variances for the independent *t*-tests. In regard to the correlational tests performed, scatterplots were examined to check for a monotonic relationship between variables.

Does social support, disengaged coping and children's behaviour impact on parents' PTSS? It was anticipated that multiple regression analyses would be performed to assess the impact of psychosocial variables on PTSS, as in keeping with previous studies in this area that examined PTSS (Bruce et al., 2011; Fuemmeler et al., 2001), and other related studies in cancer examining PTSS in parents (Greening & Stoppelbein, 2007; Norberg & Boman, 2013;). The enter method was deemed as most appropriate due to little research on what factors should be accounted for first (Clark-Carter, 2010). Relevant assumption tests were conducted before analysing this data (Clark-Carter, 2010; Field,

2009; Laerd Statistics, 2015b). Independence of residuals was examined using the Durbin-Watson statistic. Linear relationships were examined by using a scatterplot for studentized residuals against unstandardized variables and partial regression plots were used to check linearity separately. Homoscedasticity was assessed by visual inspection of the plot above and multicollinearity was checked via tolerance values. Outliers were analysed by examining the standardized residuals and the studentized deleted residuals. Leverage was assessed by inspecting the leverage values and influence was assessed by examining Cook's distance values. Finally, normality was assessed by viewing a histogram and a P-P plot of the standardized residuals.

Do parents of children with a brain tumour show significantly different levels of PTSS compared to parents of children with other medical conditions (as reported in other studies)? In order to explore the levels of PTSS in this study compared to other studies, one sample *t*-tests were planned; these types of analyses have been utilised in previous studies (Bennett, English, Rennoldson, & Starza-Smith, 2013; D'Urso, 2014; Pasterski, Mastroyannopoulou, Wright, Zucker, & Hughes, 2014). The mean of parents' PTSS in the current study was compared to a study assessing PTSS in parents of children with cancer (Norberg & Boman, 2013) and a study assessing PTSS in parents of children who had sustained an injury (Nugent et al., 2006). These studies both utilised the same scale and had similar demographics to the study sample (e.g. similar sample sizes, questionnaires completed at least 6 months post-diagnosis/event). Parametric assumption tests, as indicated above, were carried out for these analyses.

Additional Exploratory Analyses

Due to the dearth of research in this area, it was deemed important to explore the data further and assess additional relevant research questions, guided by the research to date, that could help inform future research in this area.

What is the relationship between PTSS and social support, disengaged coping, and children's behaviour? Exploratory associations between PTSS and social support, disengaged coping and behaviour were carried out using total scores and the sub-scales of the independent measures. It was anticipated that these analyses would help examine the relationship between the variables, and their sub-scales, in more detail. It was anticipated that these analyses would be conducted using Pearson's correlations and relevant assumption testing, as described above, were conducted in line with these analyses.

Are there any differences in current PTSS between parents with a history of trauma and PTSS and those without? The demographic questionnaire asked parents if they had experienced a previous trauma and if they had experienced PTSS symptoms related to this trauma. These factors have not been thoroughly investigated, however research suggests that prior psychological problems can predict post-traumatic stress (Ozer, Best, Lipsey, & Weiss, 2003) and previous life events can predict parental PTSS following a childhood cancer diagnosis (Pelcovitz et al., 1996). An exploratory analysis was planned to examine these variables using independent samples *t*-tests; the assumptions for these analyses were tested as outlined above.

Are there any differences in current PTSS between parents who have received psychological support in relation to their child's diagnosis and those who haven't? It has been reported that parental anxiety during child's treatment impacts on PTSS (Best, Streisand, Catania, & Kazak, 2001) and that early intervention and support is important in reducing PTSS in families who have experienced traumatic medical events (Kazak, 2005). Therefore, an exploratory analysis assessing the impact of psychological support received (in relation to their child's diagnosis or treatment) on PTSS was planned using an independent samples *t*-test; assumptions tests in line with this analysis were performed, as outlined above.

Are there differences in PTSS between mothers and fathers of the same child?

The study originally intended to analyse differences in parental PTSS (between parent pairs) in the empirical paper; however, this could not be conducted due to the small number of parent pairs recruited. Nevertheless, it was deemed important to conduct a tentative analysis comparing parents' PTSS. Studies have shown that mothers have higher PTSS than fathers (Baraket et al., 1997; Poder, Ljungman, & von Essen, 2008), although others report mixed evidence for this (Vrijmoet-Wiersma et al., 2008). Paired *t*-tests were planned to compare mothers and fathers PTSS, as in Kazak et al. (2004). Parametric assumptions were conducted as detailed above (using the "difference" scores between the groups).

Chapter 5. Additional Results

Missing Data

Overall, 0.16% of values were missing (N=4). MCAR was not significant (x^2 = 165.1, *df* = 164, *p* = .461), showing that cases were missing completely at random. In studies in which there is less than 5% of data missing (Tabacknick & Fidell, 2007), it is recommended that mean imputation can be conducted; this is a common method of dealing with missing data (Rubin, Witkiewits, Andre, & Reilly, 2007) and it has been suggested that it can perform as well as other missing data techniques (Peyre, Leplege, & Coste, 2011). It is of note that all measures, and sub-scales, had adequate internal reliability scores (see Appendix O) and mean and standard deviation scores were not considerably different after imputation. There was a small amount of demographic information missing and parents who had this data missing were excluded from the analysis in which this demographic data was required.

Sample Size

The study recruited 49 parents for the main analyses; this number was lower than anticipated and, therefore, some of the analyses conducted were under-powered. It was deemed that exploratory testing should still be conducted, due to including an under-researched population. The sample size recruited in the current study is also comparable to other studies using regression analyses to assess PTSS in parents of children with brain tumour (Bruce et al., 2011; Fuemmelar et al., 2001), cancer (Norberg & Boman, 2013) and other paediatric populations (Pasterski et al., 2014).

Assumption Testing

Are there significant differences in parental PTSS depending on medical/demographic variables? To examine the impact of demographic variables, it was anticipated that *t*-tests and correlational tests would be carried out. Initially it was planned that the following variables would be investigated through a correlational

analysis: time since diagnosis, time since treatment, parent age and child age. A key assumption of correlational tests is that the independent and the dependent variable show a monotonic relationship (Laerd Statistics, 2018). Child age and time since diagnosis did not show a monotonic relationship with PTSS and could not be successfully transformed. Consequently, independent sample *t*-tests were used instead and variables were dichotomized.

Normality assumptions were not met for time since treatment, as indicated by Shapiro-Wilk's test (p = .001) and the skewness value that was calculated (Z_{skew} = 2.05). The data could not be successfully transformed and, therefore, a Spearman's rank order correlation was carried out instead. Parent age was also not normally distributed, as indicated by Shapiro-Wilk's test (p = .001), and the skewness value that was calculated (Z_{skew}= 3.31), and there was one outlier. The variable could not be successfully transformed and, therefore, a Spearman's correlation was conducted instead. There was one outlier for parent sex; this outlier had a higher chance of influencing the results, due to the small and unequal sample sizes used in this analysis. In reviewing recommendations (in which guidance from Field (2009) and Laerd (2015c) were examined), it was decided that a Mann-Whitney *U* test would be carried out, which is more robust to outliers. All assumptions for the other tests were met.

Does social support, disengaged coping and children's behaviour impact on parents' PTSS? Assumptions tests for a multiple regression were carried out in line with recommendations (Field, 2009; Laerd Statistics, 2015b). Four multiple regressions were conducted and assumptions tests were completed for each regression analysis. There was independence of residuals, as assessed by the Durbin-Watson statistic, which ranged between 1.80 to 2.02 for all analyses. Linearity and homoscedasticity were met, as assessed by the scatterplots for studentized residuals against unstandardized variables and

partial regression plots. There was no evidence of multicollinearity, as assessed by tolerance values greater than 0.1. There were no studentized deleted residuals greater than ±3 standard deviations, and Cook's distance values were all above 1. In each of the regressions conducted, there were two leverage values that exceeded the cut-off recommended of up-to three times the average value (Field, 2009; Stevens, 2002). However, these were very close to the cut-off and the values did not have high Cook's distance values, and were not highlighted as outliers, therefore they were deemed not to have a high overall influence on the data. The assumption of normality was met for each regression, as assessed by the histograms and Q-Q plots.

Do parents of children with a brain tumour show significantly different levels of PTSS than parents of children with other medical conditions (as reported in other studies)? Two one-sample *t*-tests were performed to compare differences on the IES-R between parents in this sample and parents of children with other cancers and paediatric injuries. Parametric assumption tests, as indicated in the extended methodology chapter, were carried out and all assumptions for a one-sample *t*-test were met.

Additional Exploratory Analyses

What is the relationship between PTSS and social support, disengaged coping, and children's behaviour? An exploratory correlational analysis was carried out between PTSS and the primary psychosocial measures and their sub-scales; all sub-scales had good psychometric properties (as seen in Appendix O). The behaviour measure showed one outlier and was not normally distributed, as indicated by Shapiro-Wilk's test ($p < .001$) and by the skew value calculated ($Z_{skew} = 3.33$). In keeping with a positive skew, a square root transformation was applied and normality assumptions were subsequently met and there were no outliers. The emotion-focused coping scale was not normally distributed, as assessed by Shapiro-Wilk's test ($p = .028$). However, in reviewing the other normality

tests, including the histogram, QQ plot and the skewness value (Z_{skew}=-1.14) and kurtosis value ($Z_{kurtosis}$=-1.27), no deviations from normality were observed and, therefore, this was not transformed. Finally, total social support score, significant other social support score, and friends social support score were not normally distributed, as assessed by Shapiro-Wilk's test (p = .002, p < .001, p = .023, respectively), and reviewing skewness values (Z_{skew} = -3.38; Z_{skew}= -3.22; Z_{skew} = -2.14, respectively). In addition, all scales had two outliers. A square-root and reflect transformation successfully transformed social support total score and friends social support score and, following this, there were no outliers. However, significant other social support score could not be successfully transformed and, consequently, a Spearman's correlation was carried out instead for this analysis. All other measures met test assumptions for Pearson's correlations.

As seen in the Table 10, disengaged coping (r(49) = .47, p = .001) and emotion-focused disengaged coping (r(49) = .55, p < .001) were significantly correlated with PTSS. The Holm-Bonferroni method was applied and both of these analyses were still significant. Estimated effect sizes for non-significant results were all small.

Table 10

Correlations between PTSS score and psychosocial measures

Measure	r or r_s-statistic	Effect size descriptor	Significance (p value)	Holm-Bonferroni correction
Disengaged coping (total score)	r = .47	Medium	*.001**	.006
Disengaged coping (EFD)	r = .55	Large	*<.001**	.007
Disengaged coping (PFD)	r = .25	Small	.090	-
Social support (total score)[a]	r = .04	Small	.786	-
Support (SO)	r_s = .03	Small	.833	-
Support (Fri)[a]	r = -.05	Small	.737	-
Support (Fam)	r = -.16	Small	.282	-
Behaviour (total score)[a]	r = .19	Small	.189	-

Note. Disengaged coping (EFD)= Emotion-focused disengagement; Disengaged coping (PFD)= Problem-focused disengagement; Support (SO)=support from significant other; Support (Fri)= support from friends; Support (Fam)= support from family

[a]Scores have been transformed in order to meet test assumptions

*Significant correlations after Holm-Bonferroni corrections were applied

Are there any differences in current PTSS between parents with a history of trauma and PTSS and those without? Assumptions tests for independent samples *t*-tests were carried out for the impact of prior trauma history and previous PTSS; all assumptions for these tests were met. The identification of prior trauma history ($t(47) = 0.07$, $p = .947$) and self-reported PTSS related to a prior trauma ($t(47) = 0.13$, $p = .895$) were not significantly related to PTSS. Estimated effect sizes were small ($d = 0.02$ and $d = 0.04$ respectively).

Are there any differences in current PTSS between parents who have received psychological therapy in relation to their child's diagnosis and those who haven't? An independent samples *t*-test was used to explore the impact of psychological therapy, and all assumptions were met for this analysis. The experience of having psychological support did not significantly impact PTSS, $t(47) = .94$, $p = .351$. The estimated effect size was small ($d = 0.27$).

Are there differences in PTSS between mothers and fathers of the same child? Nine parent pairs took part in the study and PTSS scores of these mothers' and fathers' were compared to each other using a paired *t*-test. Assumptions tests for this test were carried out and there was one outlier detected; this was not extreme (as indicated by being less than 3 box-lengths away from the edge of the box-plot) and, as per guidance (Laerd, 2015d), the test was run with and without this and the result did not significantly change. Consequently, a paired *t*-test was carried out as planned. The analysis found that mothers had higher PTSS compared to fathers, $t(8) = 4.08$, $p = .004$; the estimated effect size was large ($d = 1.38$).

Chapter 6. Discussion and Critical Analysis

The broader aim of the thesis portfolio was to examine the emotional experiences of parents of children who have experienced a potentially life-threatening medical event; this portfolio has focused on parents of children with TBI and parents of children with a brain tumour. This chapter will elaborate on the findings in the systematic review, empirical paper and additional analyses. Results from the systematic review and empirical paper will then be discussed as a body of research together and the chapter will consider the theoretical and clinical implications of the thesis portfolio. Finally, a critical evaluation of the thesis portfolio will be provided and recommendations for future research will be made.

Systematic Review Findings

A narrative systematic review was conducted that examined parental distress following paediatric TBI. In the studies reviewed, more than half reported at least one analysis that indicated that parents of children with TBI were more distressed than parents of other children. There can be many potential complexities in caring for a child with a brain injury, such as stress related to attending to the child's needs (Aitken et al., 2004). In addition, research reports that parents have difficulty predicting the future and recovery for their child (Savage et al., 2005; Wade et al., 2001). Together, these factors might help to understand the high levels of distress reported by these parents. The systematic review also concluded that there were a variety of variables related to parents, children and families that impacted on parental distress, such as the child's brain injury severity (Hobert-Porter et al., 2015) and parents prior psychological functioning (Youngblut & Brooten, 2006, 2008). Consequently, clinicians need to be aware of a range of factors that may impact on parents' wellbeing and adjustment following paediatric TBI.

The findings of the review demonstrated that emotional responses may vary between parents and indicated that not all parents will experience poor psychological health. This is in keeping with other research reporting that not all parents are negatively impacted (Mckinlay et al., 2018; Rashid et al., 2014). In a recent systematic review on outcomes for carers of those with TBI, a handful of positive outcomes were reported, such as good family functioning (Baker, Barker, Sampson, & Martin, 2017). Indeed, Ungar (2016) reports that families show a range of different resilience patterns and coping strategies and authors point to a need for more research on families' strengths (Perlesz, Kinsella & Crowe, 1999). Furthermore, Picoraro, Womer, Kazak, and Feudtner (2014) explored post-traumatic growth in paediatric health settings and concluded that many families can find ways to benefit from traumatic experiences. Consequently, it is important to consider post-traumatic growth and factors that foster resilience in parents, as well as factors that increase their distress.

Empirical Paper Findings

Recruitment. The study recruited a similar sample size to previous studies in this area (Bruce et al., 2011; Fuemmeler et al., 2001), however it recruited fewer participants than anticipated. There are several reasons why recruitment may have been a challenge. Firstly, this is the first study in this population that has recruited parents whilst children are still in treatment. It is possible that parents might find it harder to reflect on their experiences whilst their children are still in treatment. Another possibility is that the study involved asking potentially traumatised parents to think about their experiences and, consistent with models of PTSD (Ehlers & Clark, 2000), parents who are more traumatised may show avoidance of thinking about their experiences and may be less likely to take part (Pelcovitz et al.,1996). It is also noteworthy that the study did not provide as many reminders to parents to complete the study, compared to the other two

studies in this area (Bruce et al., 2011, Fuemmeler et al., 2001), due to ethical considerations in doing this; it is possible that this impacted the recruitment rate.

Main study findings. The study findings showed a high level of PTSS in parents of children with a brain tumour, in keeping with other studies in this population (Bruce et al., 2011; Fuemmeler et al., 2001). On the PTSS measure, the intrusion scale had the highest overall mean; this is in line with findings by others whom have measured PTSS in parents of children with cancer (Bruce et al., 2011) and have asked parents to report on common symptoms (Stuber, Christakis, Houskamp, & Kazak, 1996; Yalug et al., 2008). The study assessed which factors were related to PTSS and found that only disengaged coping impacted overall PTSS. Indeed, it has been hypothesised that avoidant coping prevents change in cognitive appraisal and trauma memory, increasing threat (Ehlers & Clark, 2000). Avoidance can also hinder habituation and change in the fear network (Foa & Kazak, 1986). The mechanisms by which coping styles impact on trauma have also been studied in a sample of parents of children with cancer (Tremolada, Bonichini, Schiavo, & Pillon, 2012); it was found that reduced emotional coping predicted PTSS and that this was mediated by its impact on memory.

The study found that demographic and medical variables were not related to PTSS, after corrections for multiple comparisons were made. There is mixed evidence in the literature pertaining to these factors (Bruce, 2006; Yalug et al., 2011) and, therefore, these variables may warrant further investigation. In the regression model, social support and children's behaviour were not related to overall PTSS. As discussed in the empirical paper, there are several hypotheses about why these factors may have been non-significant. In addition to these hypotheses it is also of note that parents' scores on the social support measure were clustered towards the higher end of the scale and parents

reports of children's behaviour were clustered towards the lower end of the scale. It is possible that these distributions affected the overall significance of these factors.

Additional analyses findings. The additional analyses assessed the relationship between PTSS and the following factors: social support, disengaged coping and behaviour (including the sub-scales of these measures). The emotion-focused disengagement sub-scale and the total disengagement score correlated with PTSS, however problem-focused disengagement was not significantly correlated with PTSS. It is evident that the emotion-focused disengagement scale included statements relating to the meaning of the event, such as criticising one self. Indeed, Greening and Stoppelbein (2007) found that self-blame increases PTSD symptoms and Ehlers and Steil (1995) highlight the importance of individual appraisals of trauma. These analyses, and the research above, suggest that this particular type of coping response may put parents at the highest risk of PTSS.

An additional analysis compared mothers' and fathers' PTSS and found that mothers have significantly higher PTSS. Due to the small number of parent pairs studied this analysis is only exploratory, however it is in keeping with research which has found that mothers of children with cancer show more distress (Rodriguez et al., 2011; Yeh, 2002) and report higher PTSS than fathers (Kazak, 2005; Poder et al., 2008). One possibility is that mothers are more involved in hospital care and this may lead to further traumatisation (Vrijmoet-Wiersma et al., 2008). However, it is also possible that differences may represent general gender differences in distress (Sloper, 2000), in which women report higher levels of PTSD than men (Olff, Langeland, Draijer, & Gersons, 2007). Interestingly, in the empirical paper the analysis assessing differences in PTSS between mothers and fathers in the whole sample was not significant (although mothers scored higher than fathers). This non-significant finding could be due to the uneven sample size and power in this analysis; however, it could also suggest that differences

between mothers' and fathers' PTSS may not always be significant and further research in a larger sample is warranted.

The additional analyses found that previous traumatic experiences and PTSS did not impact on current PTSS. These findings support Manne, Duhamel, and Redd (2000) who report that lifetime prevalence of traumatic events in mothers of children with cancer is not a significant predictor of PTSS. However, other research has found that prior trauma is a significant predictor of PTSD (Brewin et al., 2000) and predicts PTSS in mothers of cancer survivors (Pelcovtiz et al., 1996). In this study the analyses were reliant on self-report of trauma and previous PTSS and, therefore, the results should be reviewed with caution. The additional analyses also found that psychological therapy, in relation to the child's diagnosis or treatment, did not impact on PTSS. This is despite research showing that parents of children with cancer can benefit from psychological support (Kreicbergs, Lannen, Onelov, & Wolfe, 2007). However, it is important to note that the length and type of psychological therapy was not recorded, and those who were seeking psychological therapy may have had higher PTSS than other parents initially. Consequently, these analyses cannot draw conclusions on the direct impact of psychological support.

Synthesis of Findings

Together the empirical paper and the systematic review demonstrated that parents of children with a brain tumour, and TBI, are at risk of psychological distress. This may not be surprising considering these parents may experience a threat to their child's life, significant uncertainty and may become aware of cognitive and behavioural changes in their child (Clark, Prior, & Kinsella, 2002; Mckinlay et al., 2014; Moore, 2005; Prasad, Swank, & Ewing-Cobbs, 2017; Snaman, Feraco, Wolfe, & Baker, 2019;Yeates et al., 2002). The findings are in keeping with a larger body of research that has found poorer

quality of life in parents of children with chronic medical conditions (Goldbeck, 2006) and poor parental wellbeing in the context of child medical illnesses (Needle et al., 2009; Pinquart, 2018; Shudy et al., 2006; Witt et al., 2010; Woolf et al., 2016).

The proportion of parental distress was particularly high in the parents studied in the empirical paper. Research suggests that post-traumatic stress is a prevalent presentation in parents of children who have been seriously ill (Ljungman et al., 2014; Rees, Gledhill, Garralda, & Nadel, 2004). Woolf et al. (2016) reviewed studies that assessed parents after paediatric illness or injury and found that between eight and 68 percent of parents experienced post-traumatic stress reactions; in this review the severity of PTSS varied between different medical conditions. More recently, Muscara et al. (2018) assessed post-traumatic stress in parents of children with serious injury or illness and found there were three separate trajectories for parents, with a small number (13%) showing consistently high PTSS. This supports the findings in this thesis portfolio that indicate that parents of children who have experienced health conditions are at risk of distress, however the trajectories and severity of distress can vary.

Both the empirical paper and the systematic review assessed factors that impact distress. The empirical paper only found one psychosocial factor that impacted PTSS (disengaged coping) and the systematic review found a variety of psychosocial and demographic variables that were reported to impact on parental distress. Whilst some research has suggested that subjective experiences are more important than objective factors (Kazak et al., 2005) and that medical factors are less likely to be associated with traumatic stress (Woolf et al., 2016), the thesis portfolio indicates that there is no clear evidence for one set of factors that impact distress. Consequently, the relative importance of demographic or medical variables, compared to psychosocial variables, remains unclear. It is possible that the heterogeneity in samples in this research field may

contribute to wide differences in research findings relating to the severity of distress and the factors that impact upon this.

Theoretical Relevance

Theories of stress and coping in caregivers. The emotional reactions of parents whom are faced with their child's critical illness can be understood using different psychological models of stress and coping. Lazarus and Folkman (1984) propose a model of stress and coping that suggests that the balance between individual appraisals of stressors and utilisation of resources is important in minimising distress. This theory remains important in stress and coping research across multiple disciplines (Biggs, Brough, & Drummond, 2017; Matthieu & Ivanoff, 2006) and has been used as a framework to understand parental experiences (Cousino & Hazen, 2013; Saloviita, Itälinna, & Leinonen, 2003). This thesis portfolio demonstrated that there are a variety of factors, or "resources", that may be influential in understanding parental distress, such as social support.

More specific theoretical models have been developed to understand parents'/caregivers' wellbeing (King, King, Rosenbaum, & Goffin, 1999; Raina et al., 2004; Wallander et al., 1989; Wallander & Varni, 1998). As discussed in the systematic review, Wallander and Varni (1998) and Wallander et al. (1989) highlight a range of risk and resistance factors that are important in understanding parent's adaption to their child's health condition. In this model factors can be those related to the child or parent and include psychological, social and medical factors. Other researchers have continued to build on this model, such as King et al. (1999), whom propose a variety of factors that can influence parental wellbeing. In this model factors that impact on parents' mental health include prognostic indicators (such as severity of disability), professional caregiving (such as the caregiving process of providing support) and mediating variables (such as social

support, coping strategies, and the child's behaviour). Raina et al. (2004) built on this further and reviewed the value of including contextual factors (such as socio-economic status), as well as caregiver strains, child characteristics, self-perception and coping styles.

The models above have been used to explore the functioning and distress of parents of ill children (Guðmundsdóttir, Guðmundsdóttir, & Elklit, 2006; Hoekstra-Weebers, Jaspers, Kamps, & Klip, 2001; Manuel, 2001; Sloper et al., 2000). These models highlight the value of not only understanding the stressful event itself (i.e. the child's illness) but the interacting factors that exist in the context around this, such as wider sources of support, caregiver demands and the family environment. The results of the systematic review were in keeping with the models in finding that there are variety of factors related to children, parents and wider family resources that can impact on psychological distress. In addition, the empirical paper explored the value of some of the variables outlined in these models, such as coping style and social support, and found that coping-style was related to parental PTSS.

Theoretical models of parental traumatic stress. There are several psychological theories of PTSD; Brewin and Holmes (2003) outline three recent theories of PTSD (emotional processing theory, dual representation theory and cognitive theory), all in which discuss the distinctive aspect of memory within PTSD. Models of PTSD, such as the cognitive model of PTSD (Ehlers & Clark, 2000) described in the empirical paper, have been used in understanding and guiding research on parental trauma in this setting (D'Urso et al., 2018; Kangas, Henry, & Bryant, 2005). In this model the nature of trauma memories, individual appraisals about the trauma and/or its sequalae and strategies to control symptoms can lead to the development and maintenance of PTSD symptoms. However, as discussed, there are debates as to if cancer can be conceptualised within a PTSD framework (Kangas et al., 2002) and it is possible that cancer-related PTSS may not

be fully explained by current models of PTSD (Bruce, 2005, 2006); therefore, it is also important to examine models that have been more specifically designed for this setting.

A model that specifically examines paediatric medical traumatic stress for children and families has been developed by Kazak et al. (2005); this model helps to provide an understanding of the impact of paediatric cancer for the whole family (Pai & Kazak, 2006) and proposes different stages that can be traumatic for families. The model proposes that families are impacted during and immediately following the event (phase one), they can then show responses related to the challenges of the medical condition and treatment (phase two) and can subsequently experience long term reactions years after treatment (phase three). This model takes into account pre-existing factors and vulnerabilities of families, as well as the characteristics and subjective experiences of the medical event. The model was recently renamed as the integrative trajectory model of paediatric medical traumatic stress (Price, Kassam-Adams, Alderfer, Christofferson, & Kazak, 2015) and underlines the different trajectories of families.

The empirical paper studied parents in the latter two stages outlined in the pediatric medical traumatic stress model (Kazak et al., 2005) and still found evidence of traumatic stress; thereby supporting the presence of PTSS in the later stages suggested by the model. The empirical paper also partly supports the model in finding that objective factors may be less important to consider in relation to PTSS (no demographic factors were found to be significant in the empirical paper). However, results from the literature as a whole are inconclusive and the importance of objective factors needs to be studied further.

Models of family adjustment. The family adjustment and adaption response model by Patterson (1988) suggests that families may cycle through repeated adjustment and adaption periods to balance demands and their own capabilities. This model suggests that parents' distress may change, based on demands and capabilities. This model may

help to understand the variation in distress documented in this thesis portfolio. Rolland's (1984) family systems illness model also takes into account processes overtime and clinicians continue to draw on this theoretical model when working with families impacted by illness (Johnston, 2015). It proposes that there are different interacting factors that impact on families, such as the time phase, onset, course and prognosis of the illness. This model considers a complex interaction of factors and helps to explore the findings in this thesis portfolio that indicate a variety of factors can impact on parents' mental wellbeing.

Clinical Implications

Implications for parents of children with TBI. The systematic review indicated that parents of children with TBI are at risk of distress; this implies that some parents would benefit from clinical services to help them adjust to their child's injury. It would be beneficial for services to consider parental needs and screen parents who may be at risk of distress, providing interventions for parents who would benefit from them. One intervention that has been researched is a web-based program for parents that involves information on problem solving, communication and managing stress (Wade, Wolfe, & Pestian, 2004). This program has been examined in parents of children with TBI and has shown improved parent child-relationships (Wade, Wolfe, & Pestian, 2004), improved parental depression (Wade, Walz, Carey, & Williams, 2008) and decreased psychiatric symptoms in parents (Wade, Carey, & Wolfe, 2006). However, a recent systematic review highlights the limited number of studies assessing parental interventions after paediatric TBI (Brown, Whittingham, Boyd, & Sofronoff, 2013) and thus research into clinical interventions in this population is still in its infancy.

Implications for parents of children with brain tumours. Although the results from the empirical paper are tentative, the high amount of trauma found in parents of children with a brain tumour underlines the need for clinicians to be aware of PTSS in this population. Indeed, Ko et al. (2008) suggest that paediatric health settings need to take a trauma-informed perspective. A service with a trauma-informed perspective can help in routinely screening for trauma, making resources available to those impacted by trauma, and strengthening protective and resilience factors (The National Child Traumatic Stress Network, n.d.).

The model of pediatric medical traumatic stress (Kazak et al., 2005) and the pediatric psychosocial preventative health model (Kazak, 2006) suggest a number of interventions at different stages. These models include normalising reactions in the early stages, to preventing or reducing PTSS in the later stages; this can be done by focusing on factors that may moderate distress, such as social support. Those showing long-term distress responses may benefit from programs such as the surviving cancer completely program (Kazak et al., 1999), outlined in the empirical paper. Parents may also benefit from trauma therapies outlined by NICE (2018) guidelines, such as trauma-focused cognitive behavioural therapy. In addition, in keeping with the finding in the empirical paper, in which disengaged coping affected PTSS, Norberg et al. (2011) have underlined the importance of research on interventions in which avoidance can be targeted, such as acceptance and commitment therapy.

Interventions for parents of children with health conditions. The thesis portfolio suggests that parents of children with medical conditions may require interventions to help to reduce or prevent psychological distress. Two recent reviews evaluated RCTs of psychological interventions (such as cognitive behavioural therapy, family therapy and problem-solving training) for parents of children with chronic illness and concluded that

there are few interventions that focus on parents and their outcomes (Eccleston, Palermo, Fisher, & Law, 2012; Eccleston, Fisher, Law, Bartlett, & Palermo, 2015). These reviews suggest that interventions are effective for some families and medical conditions, but not all, and that more research on this is warranted. In addition, it is of note that interventions may need to include all family members, as research suggests that parents can impact on children's responses and distress in medical settings (Brown, De Young, Kimble, & Kenardy, 2019). Therefore, clinical interventions may need to be facilitated whilst holding the whole family-system in mind and systemic approaches, as recommended by Rolland (1994), could be considered.

Aside from psychological therapies, it is important to assess informal models for supporting families and clinical interventions for parents more widely in health settings. Curtis, Foster, Mitchell, and Van (2016) examined models of care for families of critically ill children and reported that family-centred care can lead to positive outcomes by increasing parent satisfaction and reducing parent anxiety. Family-centred care involves promoting the wellbeing of families and aims to encourage them to become actively involved in care (Institute for Patient and Family-Centered Care, n.d.) and recognize and build on their strengths (Pettoello-Mantovani, Campanozzi, Maiuri, & Giardino, 2009). Using this model of care with families impacted by paediatric illness may help to support and increase families' resilience.

Critical Evaluation

A summary of the main strengths and weaknesses can be seen in the systematic review and the empirical paper. In the section below these strengths and weaknesses will be elaborated on and additional strengths and weaknesses will be considered.

Strengths. The empirical paper used a wider contextual framework of PTSS, therefore adapting it appropriately to the current research suggestions (Kazak et al., 2005), and noting the limitations of the diagnostic label of PTSD in this population (Kangas et al., 2002). The prevalence of distress, as well as factors that might be impacting distress, were explored in both the systematic review and the empirical paper. This meant the thesis was able to tentatively conclude on factors that might serve to increase or decrease distress, which is helpful in understanding types of clinical interventions that may be useful.

The study focused on brain tumours specifically; this area has had little research (Bruce, 2006; Bruce et al., 2011), despite the poorer quality of life of these children compared to other children with cancer (Srivastava, Pandey, & Meena, 2016) and the long-term late effects (Srivastava et al., 2016; Walter & Hilden, 2004). This not only increased the homogeneity of the sample, but it also provided a more detailed understanding of traumatic stress in these parents particularly; this has only been researched by two studies previously (Bruce et al., 2011; Fuemmeler et al., 2001). The empirical paper also helped to explore the feasibility of recruitment in this area; this included exploring a new method of recruitment in this population, using social media. Research recruiting through online platforms is proving to be a particularly important recruitment method for hard to reach populations and observational studies (King, O'Rourke, & DeLongis, 2014; Topolovec-Vranic & Natarajan, 2016). This study was able to document the effectiveness of this approach for this population, which can be used to help inform future studies.

Weaknesses. The main weakness of the empirical paper was the size of the sample recruited. Recruiting parents of children with cancer is notoriously difficult (Akard et al., 2015) and this is why a number of recruitment strategies were used. Despite

this, the recruitment rate was 20%; this is less than the two other studies in this area who recruited 39% (Fuemmeler et al., 2001) and 37% (Bruce et al., 2011) and, therefore, the sample may not have been representative of the population. It is possible that this impacted on the prevalence scores reported; for example, it is possible that parents who felt more distressed might have had more motivation to participate in the study, thereby inflating PTSS scores. However, Pelcovitz et al. (1996) studied parents of children with cancer and suggested that individuals who didn't take part were more likely to have higher PTSD. Considering this, it is not clear if those who didn't take part had significantly higher or lower PTSS scores. Nevertheless, the small sample size meant that some of the analyses were under-powered and, therefore, the study may not have been able to detect the significance of some of the factors studied.

The study set out to recruit and include fathers but recruited a relatively small number (N=14). It was also evident that the studies in the systematic review were more likely to include a larger sample of mothers, compared to fathers. The imbalances between the number of mothers and fathers recruited in these samples might be reflective of research that has suggested that fathers are more difficult to recruit (Phares, 1995) and are less likely to take part than mothers (MacFadyen et al., 2011). Unfortunately, this meant that the thesis portfolio was only partly reflective of fathers' experiences. It was also evident that the study recruited largely a sample of white British parents, and parents whose occupations were considered as "managerial" or "professional"; consequently, the sample may have under-represented minority ethnic groups and parents with other occupational backgrounds. It is also of note that 31% of parents in the empirical study reported previous PTSS. Although the study could not conclude if they had a diagnosis of PTSD, the impact of previous trauma may have been higher in this sample than in the general population (with a rate of 6% of life-time PTSD; Frans, Rimmö, Åberg, &

Fredrikson, 2005) and, consequently, this may have impacted on the generalisability of the results.

In relation to the measures used in the study, scores on the behavioural measure were clustered towards the lower end of the scale; one hypothesis for this could be that children's behaviour was not a significant concern for these parents or, alternatively, the measure may not have been sensitive to behavioural problems experienced by these children. Initially, the study set out to use the Strengths and Difficulties Questionnaire (Goodman, 1997), which has been used in this population (Bruce, 2005), however this was not freely available for online use. Instead the ODD scale was used (Burns et al., 2001; Burns et al., 2015), and this is the first time the measure has been used in this exact setting. Although valid in other settings (Burns et al., 2008) and recommended as a "free" measure (Beidas et al., 2015), the findings might indicate that this measure may not be sensitive in rating children's behaviour in this setting; this is a useful finding for future studies in this area. As discussed above, the social support measure indicated that many parents reported high levels of support. The original measure developed also found that many participants were categorised as having high levels of support (Canty-Mitchell & Zimet, 2000; Zimet et al., 1988). Therefore, a potential limitation of this measure is that it may be weak in differentiating between different levels of social support.

Future Research

In this thesis portfolio it was evident that research assessing parents of children with TBI, and parents of children with cancer, often included heterogenous samples and it would be beneficial to recruit more homogeneous samples. This would allow for further exploration of the particular difficulties faced by parents. It would also allow for a better understanding of the course and prognosis of the illness and the interactions between this and parents' adjustment, as outlined as important by Rolland (1994). Indeed, it has been

suggested that future research should address family needs within the specific context of that illness (Golfenshtein, Srulovici, & Medoff-Cooper, 2016).

In both the empirical paper and the systematic review, it was identified that many studies recruited a sample consisting largely of mothers. It is important that research continues to try to recruit fathers, and engage them in research, so that services can address both parents' needs. Kazak et al. (2004) reports that 37% of families have a family member with PTSD at some point after the cancer diagnosis, therefore it is important to not focus on one parent. Similar to this, it is important to continue to explore distress in other members of the family, such as siblings, whom have also shown to have a high rate of distress (Long et al., 2018; Sharpe & Rossiter, 2002). It is also of note that many research studies reviewed in this thesis portfolio included samples of parents in the US. Due to differences between countries health-care systems and models related to cancer care (Jeffard et al., 2013) and TBI care (Cnossen et al., 2016) it would be valuable to conduct more research to explore parents' wellbeing in UK samples to increase homogeneity in samples and explore the interactions between parents and particular health-care systems.

Many studies included in the review and the empirical paper were cross-sectional. Theories suggest that parents' adjustment and adaption can change over-time (Patterson, 1988; Price et al., 2015; Rolland, 1994), therefore it would be helpful to track this using longitudinal research. It would also be valuable to conduct more qualitative studies in this area. Researchers continue to write about the individuality and complexity of families impacted by paediatric illness (Kazak, 1997; Rolland, 1994) and, therefore, an exploration of their lived experience would continue to add to the literature and help to provide a further understanding of how services might be able to support families.

It is of note that whilst research examining post-traumatic stress in parents of children with cancer is growing, PTSS has not been assessed in parents of children with TBI. Considering some of the experiences of parents with children with TBI noted in the systematic review and the high prevalence of trauma found in the empirical paper, it might be valuable for future research to study PTSS in parents of children with TBI. In future research that assesses trauma responses in medical settings, the model of pediatric traumatic medical stress (Kazak et al., 2005) is helpful in outlining family responses over-time. However, the model is limited in not addressing and outlining key mechanisms for the development and maintenance of post-traumatic stress. Consequently, further research developing this model, and current models of PTSD, for parents of children with health conditions would be beneficial. As models are refined it would be valuable to use these models to guide research as it has been acknowledged that research in this area has lacked a theoretical structure (Bruce, 2006; Drotar, 1997) and studies have not had big enough sample sizes to test particular theoretical models (Sloper, 2000).

Considering the research that has been reviewed in this area that has suggested that parents of ill children may be at risk of distress, it would be beneficial for future research to focus on designing and evaluating interventions for these parents. In paediatric illness it has been reported that there is little research on what interventions are effective for parents (Eccleston et al., 2012). This is an area that would benefit from continued research so that services are aware of how they can help families with distress related to their child's illness.

As highlighted above, recruitment for the empirical paper was lower than expected. Future studies should explore the acceptability of recruiting in this population and would benefit on gaining parents feedback on barriers to getting involved in research. Recommendations from the empirical study suggest that future research may benefit from

being online to help enhance recruitment. It may also be beneficial for future studies to use a longer recruitment period and more research sites. Finally, it might be helpful to provide parents with extra prompts (e.g. sending more research packs if parents haven't responded), as did Bruce et al. (2011), however, the ethical implications and acceptability of this for parents would need to be explored further.

Conclusions

The thesis portfolio aimed to further understand the experience of parents of children with TBI and parents of children with a brain tumour. The findings of the thesis portfolio suggest that these parents are vulnerable to experiencing emotional distress and, therefore, clinical services would benefit from supporting the wider family. The thesis portfolio indicates that factors that impact on parents' responses may vary and, consequently, clinicians need to take a person-centred approach. This research area would benefit from further studies exploring the presentation of distress in parents of children with serious illnesses. Furthermore, it would be valuable for future research studies to design and evaluate clinical interventions for parents of children with health conditions.

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Appendices

Appendix A

Author guidelines for journal of the International Neuropsychological Society (see critical review)

Instructions for contributors

Aims and Scope

The *Journal of the International Neuropsychological Society* is the official journal of the International Neuropsychological Society, an organization of over 4,500 international members from a variety of disciplines. The *Journal of the International Neuropsychological Society* welcomes original, creative, high quality research papers covering all areas of neuropsychology. The focus of articles may be primarily experimental, applied, or clinical. Contributions will broadly reflect the interest of all areas of neuropsychology, including but not limited to: development of cognitive processes, brain-behavior relationships, adult and pediatric neuropsychology, neurobehavioral syndromes (such as aphasia or apraxia), and the interfaces of neuropsychology with related areas such as behavioral neurology, neuropsychiatry, genetics, and cognitive neuroscience. Papers that utilize behavioral, neuroimaging, and electrophysiological measures are appropriate.

To assure maximum flexibility and to promote diverse mechanisms of scholarly communication, the following formats are available in addition to a *Regular Research Article: Brief Communication* is a shorter research article; *Rapid Communication* is intended for "fast breaking" new work that does not yet justify a full length article and is placed on a fast review track; *Case Report* is a theoretically important and unique case study; *Critical Review* and *Short Review* are thoughtful considerations of topics of importance to neuropsychology and include meta-analyses; *Dialogue* provides a forum for publishing two distinct positions on controversial issues in a point-counterpoint format; *Special Issue* and *Special Section* consist of several articles linked thematically; *Letter to the Editor* responds to recent articles published in the *Journal of the International Neuropsychological Society*; and *Book Review,* which is considered but is no longer solicited.

Originality and Copyright

To be considered for publication in the *Journal of the International Neuropsychological Society*, a manuscript cannot have been published previously nor can it be under review for publication elsewhere. Papers with multiple authors are reviewed with the assumption that all authors have approved the submitted manuscript and concur with its submission to the *Journal of the International Neuropsychological Society.*

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Disclosure

Potential conflicts of interest include funding sources for the reported study (e.g., a test validation study financially supported by a test publisher, a study supported by an insurance company), personal or family financial interest in a test or product or with a company that publishes a test that is being investigated in the manuscript or competes with a test that is being investigated in the manuscript. Other conflicts include employment, consultancies, stock ownership or medicolegal work. For the latter, information about whether the author's medicolegal work is largely for one side should be reported. This list of potential conflicts is not all inclusive, and it is the responsibility of each author to ensure that all of their "potential conflicts" are reported in the Acknowledgment section of the paper.

Disclosure pertains to all authors. It is the corresponding author's ethical responsibility to explicitly check with each of his/her co-authors to ensure that any real or apparent conflict of interest is appropriately disclosed. Authors should err on the side of full disclosure, and if authors are uncertain about what constitutes a relevant conflict, they should contact the Editorial Office (jins@cambridge.org). The intent of this disclosure is not to prevent an author with a significant financial or other relationship from publishing their work in the Journal of the International Neuropsychological Society, but rather to provide readers with adequate information to form their own judgments about the work.

Compliance with institutional research standards for animal or human research (including a statement that the research was completed in accordance with the Helsinki Declaration should be included in the methods section of the manuscript.

Manuscript Submission and Review

The Journal of the International Neuropsychological Society uses online submission and peer review. Paper submissions are not accepted. Authors who are not able to submit their manuscripts online are asked to contact the editorial office at: jins@cambridge.org.

The website address for submissions is: http://mc.manuscriptcentral.com/jins. Complete instructions are provided on the website.

Prior to online submission, please consult http://www.nlm.nih.gov/mesh/ for 6 keywords or mesh terms that are different from words in the title. Accurate mesh terms will increase the probability that your manuscript will be identified in online searches. Please follow the instructions carefully to avoid delays. The menu will prompt the author to provide all necessary information, including the manuscript category, the corresponding author including postal address, phone and fax numbers, and e-mail address, and suggested reviewers.

The website will automatically acknowledge receipt of the manuscript and provide a manuscript reference number. The Editor-in-Chief will assign the manuscript for review to an action editor and at least two other reviewers. Every effort will be made to provide the author with a review within 6 to 10 weeks of manuscript assignment. Rapid Communications will be reviewed within 4 weeks. If the Editor requests that revisions be made to a manuscript before publication, a maximum of 3 months will be allowed for preparation of the revision, except in unusual circumstances.

Manuscript Length

In order to increase the number of manuscripts that can be published in the *Journal of the International Neuropsychological Society*, please adhere to the following length requirements. Please provide a word count on the title page for the abstract and manuscript (not including abstract, tables, figures, or references). Manuscripts will be returned if they exceed length requirements.

Regular Research Article: Maximum of 5,000 words (not including abstract, tables, figures, or references) and a 250 word abstract. Regular Research Articles are original, creative, high quality papers covering all areas of neuropsychology; focus may be experimental, applied or clinical. Brief and Rapid Communications: Maximum of 2,500 words (not including abstract, tables, figures, or references) and a 200 word abstract, with a maximum of two tables or two figures, or one table and one figure, and 20 references. Brief and Rapid Communications are shorter research articles.

Brief Communication/Rapid Communication: Maximum of 2,500 words (not including abstract, tables, figures, or references) and a 200 word abstract, with a maximum of two tables or two figures, or one table and one figure, and 20 references. Brief and Rapid Communications are shorter research articles.

Case Report: Maximum of 3,500 words with an informative literature review (not including abstract, tables, figures, or references) and a 200 word abstract. Neurobehavioral Grand Rounds are unique case studies that make a significant theoretical contribution.

Critical Review: Maximum of 7,000 words (not including abstract, tables, figures, or references) and a 250 word abstract. Critical Reviews will be considered on any important topic in neuropsychology. Quantitative meta-analyses are encouraged. Critical Reviews must be preapproved by the Editor-in-Chief. For consideration, please e-mail your abstract to jins@cambridge.org.

Short Review: Maximum of 2,500 words (not including abstract, tables, figures, or references) and a 150 word abstract. Short Reviews are conceptually oriented snapshots of the current state of a research area by experts in that area. Short Reviews must be preapproved by the Editor-in-Chief. For consideration, please e-mail your abstract to jins@cambridge.org.

Dialogue: Maximum of 2,000 words for each segment (not including abstract, tables, figures, or references) and a 150 word abstract, with a maximum of two tables or two figures, or one table and one figure and 20 references. Dialogues provide a forum for two distinct positions on controversial issues in a point counterpoint form. Dialogues must be preapproved by the Editor-in-Chief. For consideration, please e-mail your abstract to jins@cambridge.org.

Special Issue/Special Section: Maximum of 5,000 words (not including abstract, tables, figures, or references) and a 250 word abstract for each article (same as Regular Research Articles). Symposia consist of several thematically linked research articles which present empirical data. Symposia must be pre-approved by the Editor-in- Chief. For consideration, e-mail your proposal to jins@cambridge.org to receive prior approval.

Letter to the Editor: Maximum of 500 words (not including table, figure, or references) with up to five references and one table or one figure. Letters to the Editor respond to recent articles published in the *Journal of the International Neuropsychological Society*.

Book Review: Maximum of 1000 words in length. Include name and affiliations, a title for the review, the author(s)/editor(s), title, publisher, date of publication, number of pages and price. For consideration, e-mail jins@cambridge.org.

Manuscript Preparation and Style

The entire manuscript should be typed double-spaced throughout using a word processing program. Unless otherwise specified, the guideline for preparation of manuscripts is the *Publication Manual of the American Psychological Association (6th edition)* except for references with 3 or more authors (see References section). This manual may be ordered from: APA Order Dept., 750 1st St. NE, Washington, DC 20002-4242, USA.

Pages should be numbered sequentially beginning with the Title Page. The Title Page should contain the full title of the manuscript, the full names and institutional affiliations of all authors; mailing address, telephone and fax numbers, and e-mail address for the corresponding author; and the word count for the abstract and manuscript text (excluding title page, abstract, references, tables, and figures). At the top right provide a short title of up to 45 characters preceded by the lead author's last name. Example: Smith-Memory in Parkinson's Disease. This running head should be repeated at the top right of every following page.

Page 2 should include an Abstract and a list of at least six keywords or mesh terms. Note: structured abstracts must be included with papers submitted after January 1, 2014. A structured abstract must include four header labels: Objective, Method, Results, and Conclusions. A total of six mesh terms (http://www.nlm.nih.gov/mesh/) or keywords should be provided and should not duplicate words in the title.

The full text of the manuscript should begin on page 3. For scientific articles, including *Regular Research Articles*, *Brief Communications*, *Rapid Communications*, and *Symposia*, the format should include a structured Abstract, Introduction, Method, Results, and Discussion. This should be followed by Acknowledgments, References, Tables, Figure Legends, Figures, and optional Appendices and Supplemental Material.

The use of abbreviations, except those that are widely used, is strongly discouraged. They should be used only if they contribute to better comprehension of the manuscript. Acronyms should be spelled out at first mention. Metric system (SI) units should be used.

Appendices and Supplemental Materials may be submitted. Appendices include material intended for print and should be included with the manuscript file. Supplementary material will appear only online and should be submitted as a separate file. Supplementary material is replicated as-is.

The Acknowledgements Section should include a disclosure of conflicts of interest (see above) and all sources of financial support for the paper. In documenting financial support, please provide details of the sources of financial support for all authors, including grant numbers. For example, "This work was supported by the National Institutes of Health (grant number XXXXXXX)". Multiple grant numbers should be separated by a comma and space and where research was funded by more than one agency, the different agencies should be separated by a semicolon with "and" before the final funding agency. Grants held by different authors should be identified using the authors' initials. For example, "This work was supported by the Wellcome Trust (A.B., grant numbers XXXX, YYYY), (C.D., grant number ZZZZ); the Natural Environment Research Council (E.F.,

grant number FFFF); and the National Institutes of Health (A.B., grant number GGGG), (E.F., grant number HHHH)."

Tables and Figures should be numbered in Arabic numerals. Figures should be numbered consecutively as they appear in the text. Figures should be twice their intended final size and authors should do their best to construct figures with notation and data points of sufficient size (recommended ≥ 300 dpi) to permit legible photo reduction to one column of a two-column format. Please upload figure(s) in either a .doc, .jpeg, .tiff, or .pdf format. There is no additional cost for publishing color figures. The approximate position of each table and figure should be provided in the manuscript with call-outs: [INSERT TABLE 1 HERE]. Tables and figures should be on separate pages. Tables should have short titles and all figure legends should be on separate pages. All tables and figures must have in-text citations in order of appearance.

Figures submitted in color will appear online in color, but all figures will be printed in black and white unless authors specify during submission that figures should be printed in color, for which there may be a fee. There is no additional cost for publishing color figures in the print version of the journal for corresponding authors who are INS members. For non-members, the cost for publishing color figures in print version of the journal will be $320 per figure with a cap of $1600 per article.

References should be consistent with the *Publication Manual of the American Psychological Association (6th Edition)*. In-text references should be cited as follows: "...Given the critical role of the prefrontal cortex (PFC) in working memory (Cohen et al., 1997; Goldman-Rakic, 1987; Perlstein et al., 2003a, 2003b)..." with multiple references in alphabetical order. Another example: "...Cohen et al. (1994, 1997), Braver et al. (1997), and Jonides and Smith (1997) demonstrated..."

References cited in the text with two authors should list both names. References cited in the text with three, four, or five authors, list all authors at first mention; with subsequent citations include only the first author's last name followed by et al. References cited in the text with six or more authors should list the first author et al. throughout. In the reference section, for works with up to seven authors, list all authors. For eight authors or more, list the first six, then ellipses followed by the last author's name. Examples of the APA reference style are as follows:

Online/Electronic Journal Article with DOI: Dikmen, S., Machamer, J., Fann, J. & Temkin, N. (2010). Rates of symptom reporting following traumatic brain injury. *Journal of the International Neuropsychological Society*, 16, 401-411. doi:10.1017/S1355617710000196

Scientific Article: Giovannetti, T., Britnell, P., Brennan, l., Siderowf, A., Grossman, M., Libon, D.J., Seidel, G.A. (2012). Everyday action impairment in Parkinson's disease dementia. *Journal of the International Neuropsychological Society*, 18, 787-798.

Book: Lezak, M.D., Howieson, D.B., Bigler, E.D., Tranel, D. (2012). *Neuropsychological Assessment*. New York: Oxford University Press.

Book Chapter: Mahone, E.M. & Slomine, B.S. (2008). Neurodevelopmental disorders. In J.E.Morgan, & J.H. Ricker (Eds.), *Textbook of Clinical Neuropsychology* (pp. 105-127). New York:Taylor & Francis.

Report at a Scientific Meeting: Weintraub, S. (2012, June). Profiles of dementia: Neuropsychological, neuroanatomical and neuropathologic phenotypes. International Neuropsychological Society, Oslo, Norway.

Manual, Diagnostic Scheme, etc.: American Psychiatric Association (1994). *Diagnostic and Statistical Manual of Mental Disorders* (4th ed.). Washington, DC: American Psychiatric Association Press.

Appendix B

Quality criteria

Original Downs and Black scale (1998) can be accessed in appendix of original paper:

Downs, S. H., & Black, N. (1998). The feasibility of creating a checklist for the assessment of the methodological quality both of randomised and non-randomised studies of health care interventions. *Journal of Epidemiology & Community Health, 52*(6), 377-384. doi: 10.1136/jech.52.6.377

The following papers were also reviewed for guidance:

Von Elm, E., Altman, D. G., Egger, M., Pocock, S. J., Gøtzsche, P. C., Vandenbroucke, J. P., & Strobe Initiative. (2007). The Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) statement: guidelines for reporting observational studies. *PLoS Medicine, 4*(10), e296. doi: 10.1007/s10826-013-9781-7

Kmet, L. M., Lee, R. C., & Cook, L. S. (2004). *Standard quality assessment criteria for evaluating primary research papers from a variety of fields*. Alberta: Alberta Heritage Foundation for Medical Research. Retrieved from https://www.ihe.ca/advanced-search/standard-quality-assessment-criteria-for-evaluating-primary-research-papers-from-a-variety-of-fields

Modified version utilised in the systematic review

Quality review: Questions 1-6= reporting quality Questions 7-12=methodological quality	Yes (1)	No (0)
1.Are the hypotheses/aims/objectives of the study clearly described?		
2.Are the main outcomes, including the emotional distress measure, to be measured clearly described in the Introduction or Methods section? *If the main outcomes are first mentioned in the Results section, the question should be answered no. If an outcome is mentioned but not described answer no.*		
3. Are the characteristics of the patients included in the study clearly described? *In cohort studies and trials, inclusion and/or exclusion criteria should be given. In case-control studies, a case-definition and the source for controls should be given.*		
4. Are the main findings of the study clearly described? *Simple descriptive data should be reported for all major findings and hypotheses, including the hypotheses related to the emotional distress measure.*		
5. Does the study provide estimates of the random variability in the data for the main outcomes, including the emotional distress measure? *In non-normally distributed data the interquartile range of results should be provided. In normally distributed data the mean, standard error, standard deviation or confidence intervals of the main outcome measures should be reported.*		

6. Have actual probability values been reported? *Have actual probability values been reported for significant and non-significant results (e.g.0.035 rather than <0.05) for all the main outcomes, including the emotional distress measure, except where the probability value is less than 0.001?*		
7. Has missing data been identified and addressed where appropriate? *Yes if reference to missing data and how this was addressed. If reported missing data but did not say how this was dealt with, answer no. If did not report if there was missing data or not answer no.*		
8. Have confounding variables been identified and controlled for, where appropriate? *For example, if comparing groups have they examined significant differences in demographic variables and controlled for them/ considered controlling where appropriate. Yes if a list of potential confounders is provided and included in statistical analysis, where appropriate. Answer no if no potential confounders have been identified or considered or they have been identified but not controlled in main analyses with no justification.*		
9. Were the subjects asked to participate in the study representative of the population from which they were recruited? *The study must identify the source population for patients and describe how the patients were selected. Patients would be representative if they appear to have been recruited in a non-biased way and represent an effort to recruit a representative sample of the inclusion criteria that the study set out to recruit.*		
10. Is the data free from "data dredging" and if any of the results of the study were based on "data dredging", was this made clear? *Any analyses that had not been planned at the outset of the study should be clearly indicated. If no retrospective unplanned subgroup analyses were reported, then answer yes.*		
11. Were the statistical tests used to assess parent's emotional distress appropriate? *The statistical techniques used must be appropriate to the data. For example, nonparametric methods should be used for small sample sizes. Where little statistical analysis has been undertaken but where there is no evidence of bias, the question should be answered yes. If the distribution of the data (normal or not) is not described it must be assumed that the estimates used were appropriate and the question should be answered yes.*		
12. Were the main outcome measures for parental emotional distress used accurate (valid and reliable)? *For studies that have used a well-known validated instrument the question should be answered as yes. Where the validity/reliability of the outcome measures are clearly described (eg. Inter-reliability reported), the question should be answered yes. For studies which refer to other work or that demonstrates the outcome measures are accurate (references validity of measures), the question should be answered as yes.*		

Appendix C

Reference list of excluded articles

Articles in which full texts were examined and then subsequently excluded (N=30)

Subheadings for why articles were excluded are given and, in some cases, an additional explanation is given below the references in italics

- Adult TBI sample/mixed sample including 'adolescents' over age 18

Harris, J. K., Godfrey, H. P., Partridge, F. M., & Knight, R. G. (2001). Caregiver depression following traumatic brain injury (TBI): a consequence of adverse effects on family members?. *Brain Injury, 15*(3), 223-238. doi: 10.1080/02699050010004040

Hawley, C. A., Ward, A. B., Magnay, A. R., & Long, J. (2003). Parental stress and burden following traumatic brain injury amongst children and adolescents. *Brain Injury, 17*(1), 1-23. doi: 10.1080/0269905021000010096

(Used up to age 21)

Kersel, D. A., Marsh, N. V., Havill, J. H., & Sleigh, J. W. (2001). Psychosocial functioning during the year following severe traumatic brain injury. *Brain injury, 15*(8), 683-696. doi: 10.1080/02699050121354

Kreutzer, J. S., Rapport, L. J., Marwitz, J. H., Harrison-Felix, C., Hart, T., Glenn, M., & Hammond, F. (2009). Caregivers' well-being after traumatic brain injury: a multicenter prospective investigation. *Archives of Physical Medicine and Rehabilitation, 90*(6), 939-946. doi:10.1080/026990596124296

Mangeot, S., Armstrong, K., Colvin, A. N., Yeates, K. O., & Taylor, H. G. (2002). Long-term executive function deficits in children with traumatic brain injuries: Assessment using the Behavior Rating Inventory of Executive Function (BRIEF). *Child Neuropsychology, 8*(4), 271-284. doi: 10.1076/chin.8.4.271.13503

(Used up to age 19)

Marsh, N. V., Kersel, D. A., Havill, J. H., & Sleigh, J. W. (1998). Caregiver burden at 6 months following severe traumatic brain injury. *Brain injury, 12*(3), 225-238. doi: 10.1080/026990598122700.

Perlesz, A., & O'loughlan, M. (1998). Changes in stress and burden in families seeking therapy following traumatic brain injury: a follow-up study. *International Journal of Rehabilitation research, 21*(4), 339-354

Perrin, P. B., Stevens, L. F., Sutter, M., Hubbard, R., Sosa, D. M. D., Jove, I. G. E., & Arango-Lasprilla, J. C. (2013). Exploring the connections between traumatic brain injury caregiver mental health and family dynamics in Mexico City, Mexico. *PM&R, 5*(10), 839-849. doi: 10.1016/j.pmrj.2013.05.018

- No separate parental distress measure analysed or measure too broad and included general stresses

Hawley, C. A. (2012). Self-esteem in children after traumatic brain injury: An exploratory study. *NeuroRehabilitation, 30*(3), 173-181.

(No separate parental distress measure)

Hermans, E., Winkens, I., Winkel-Witlox, S. T., & van Iperen, A. (2012). Caregiver reported problems of children and families 2–4 years following rehabilitation for pediatric brain injury. *NeuroRehabilitation, 30*(3), 213-217. doi: 10.3233/NRE-2012-0747.

(Looked at family environment and did not used this as an outcome measure)

Karver, C. L., Kurowski, B., Semple, E. A., Stancin, T., Taylor, H. G., Yeates, K. O., ... & Wade, S. L. (2014). Utilization of behavioral therapy services long-term after traumatic brain injury in young children. *Archives of Physical Medicine and Rehabilitation*, *95*(8), 1556-1563. doi: 10.1016/j.apmr.2014.03.030

(No separate parental distress measure)

Mather, F. J., Tate, R. L., & Hannan, T. J. (2003). Post-traumatic stress disorder in children following road traffic accidents: A comparison of those with and without mild traumatic brain injury. *Brain injury*, *17*(12), 1077-1087. doi: 10.1080/0269905031000114045

(No separate parental distress measure)

Mckinlay, A., Albicini, M., & Than, M. (2017). Preinjury characteristics of children with mild traumatic brain injury : Is " other injury " an appropriate comparison group "? *Journal of Clinical and Experimental Neuropsychology*, *0*(0), 1–7. doi: 10.1080/13803395.2017.1342771

(Measured parental distress which included general stress and factors related to the general parenting role)

Montgomery, V., Oliver, R., Reisner, A., & Fallat, M. E. (2002). The effect of severe traumatic brain injury on the family. *Journal of Trauma and Acute Care Surgery*, *52*(6), 1121-1124.

(No separate distress measure analysed separately using significance tests)

Schmidt, A. T., Hanten, G. R., Li, X., Orsten, K. D., & Levin, H. S. (2010). Emotion recognition following pediatric traumatic brain injury: Longitudinal analysis of emotional prosody and facial emotion recognition. *Neuropsychologia*, *48*(10), 2869-2877. doi: 10.1016/j.neuropsychologia.2010.05.029.

(Used a broader measure of life stressors)

Sokol, D. K. (1996). Behavioural adjustment and parental stress associated with closed head injury in children. *Brain injury*, *10*(6), 439-451. doi: 10.1080/026990596124296

(Used a general stress measure)

Yeates, K. O., Swift, E., Taylor, H. G., Wade, S. L., Drotar, D., Stancin, T., & Minich, N. (2004). Short-and long-term social outcomes following pediatric traumatic brain injury. *Journal of the International Neuropsychological Society*, *10*(3), 412-426. doi: 10.1017/S1355617704103093

(Used a broader measure not related to distress)

- Parental distress not used as an outcome measure (studies examined impact of parental distress on other variables)

Catroppa, C., Hearps, S., Crossley, L., Yeates, K., Beauchamp, M., Fusella, J., & Anderson, V. (2017). Social and Behavioral Outcomes following Childhood Traumatic Brain Injury: What Predicts Outcome at 12 Months Post-Insult? *Journal of Neurotrauma, 34*(7), 1439-1447. doi: 10.1089/neu.2016.4594

Kinsella, G., Ong, B., Murtagh, D., Prior, M., & Sawyer, M. (1999). The role of the family for behavioral outcome in children and adolescents following traumatic brain injury. *Journal of Consulting and Clinical Psychology*, *67*(1), 116. doi: 10.1037/0022-006X.67.1.116

McNally: McNally, K. A., Bangert, B., Dietrich, A., Nuss, K., Rusin, J., Wright, M., ... & Yeates, K. O. (2013). Injury versus noninjury factors as predictors of postconcussive symptoms following mild traumatic brain injury in children. *Neuropsychology*, *27*(1), 1**.** doi:10.1037/a0031370

Olsson, K. A., Lloyd, O. T., LeBrocque, R. M., McKinlay, L., Anderson, V. A., & Kenardy, J. A. (2013). Predictors of child post-concussion symptoms at 6 and 18 months following mild traumatic brain injury. *Brain injury*, *27*(2), 145-157. doi: 10.3109/02699052.2012.729286

Wade, S. L., Cassedy, A., Walz, N. C., Taylor, H. G., Stancin, T., & Yeates, K. O. (2011). The relationship of parental warm responsiveness and negativity to emerging behavior problems following traumatic brain injury in young children. *Developmental Psychology*, *47*(1), 119. doi: 10.1037/a0021028

- Correlations or percentages used to examine outcome variables

Aitken, M. E., McCarthy, M. L., Slomine, B. S., Ding, R., Durbin, D. R., Jaffe, K. M., ... & MacKenzie, E. J. (2009). Family burden after traumatic brain injury in children. *Pediatrics*, *123*(1), 199-206. doi: 10.1542/peds.2008-0607

(Only used percentages)

Josie, K. L., Peterson, C. C., Burant, C., Drotar, D., Stancin, T., Wade, S. L., ... & Taylor, H. G. (2008). Predicting family burden following childhood traumatic brain injury: a cumulative risk approach. *The Journal of Head Trauma Rehabilitation*, *23*(6), 357. doi: 10.1097/01.HTR.0000341431.29133.a8

Peterson, R. L., Kirkwood, M. W., Taylor, H. G., Stancin, T., Brown, T. M., & Wade, S. L. (2013). Adolescents' internalizing problems following traumatic brain injury are related to parents' psychiatric symptoms. *The Journal of Head Trauma Rehabilitation*, *28*(5), E1. doi: 10.1097/HTR.0b013e318263f5ba

Youngblut, J. M., Brooten, D., & Kuluz, J. (2005). Parents' reactions at 24–48 hours after a preschool child's head injury. *Pediatric critical care medicine: a journal of the Society of Critical Care Medicine and the World Federation of Pediatric Intensive and Critical Care Societies*, *6*(5), 550.

- Mixed sample of injuries (TBI mixed with other injury groups)

Daviss, W. B., Racusin, R., Fleischer, A. M. Y., Mooney, D., Ford, J. D., & McHUGO, G. J. (2000). Acute stress disorder symptomatology during hospitalization for pediatric injury. *Journal of the American Academy of Child & Adolescent Psychiatry*, *39(*5), 569-575. doi: 10.1097/00004583-200005000-00010

Ostrowski, S. A., Christopher, N. C., & Delahanty, D. L. (2006). Brief report: the impact of maternal posttraumatic stress disorder symptoms and child gender on risk for persistent posttraumatic stress disorder symptoms in child trauma victims. *Journal of Pediatric Psychology*, *32*(3), 338-342. doi: 10.1093/jpepsy/jsl003

- Included control group, with TBI group, in overall analysis of factors that impact distress

Wade, S. L., Stancin, T., Taylor, H. G., Drotar, D., Yeates, K. O., & Minich, N. M. (2004). Interpersonal stressors and resources as predictors of parental adaptation following pediatric traumatic injury. *Journal of Consulting and Clinical Psychology*, *72*(5), 776. doi: 10.1037/0022-006X.72.5.776

Wade, S. L., Taylor, H. G., Drotar, D., Stancin, T., Yeates, K. O., & Minich, N. M. (2003). Parent-Adolescent Interactions After Traumatic Brain Injury: Their Relationship to Family Adaptation and Adolescent Adjustment. *The Journal of Head Trauma Rehabilitation*, *18*(2), 164-176. doi: 10.1097/00001199-200303000-00007

Note

A refresh search took place in August 2018. One potentially relevant new article was found, however on review this was excluded, due to using a parenting distress measure that measured distress in relation to the parenting role:

Gagner, C., Landry-Roy, C., Bernier, A., Gravel, J., & Beauchamp, M. H. (2018). Behavioral consequences of mild traumatic brain injury in preschoolers. *Psychological medicine*, *48*(9), 1551-1559. doi: 10.1017/S0033291717003221

Appendix D

Author guidelines for Children's Health Care

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Updated 15-01-2019

Appendix E

Information sheet hospital (final version)

Version 4 September 2018

University of East Anglia

Examining the wellbeing of parents of children who have been diagnosed with a brain tumour

My name is Briony Westgate and I am a Trainee Clinical Psychologist based at the University of East Anglia (UEA). I would like to invite you to take part in a research study that is being conducted. Before you can take part, it is important that you read this information sheet; please take your time to read it carefully and contact me if you would like further information or have any questions (contact details are at the end). You do not have to make any immediate decisions about taking part.

What is the purpose of the project?

This project aims to looks at parents' wellbeing following their child's brain tumour diagnosis. The study will look at post-traumatic stress symptoms and look at what factors might contribute to this. Post-traumatic stress symptoms can include having nightmares, trouble concentrating, and feeling numb, amongst other symptoms. There is little research into what factors might impact this in parents of children with a brain tumour and this research aims to increase our understanding of what may contribute to this.

Can I take part?

We are asking parents of children aged 4-16 years who have been diagnosed with a brain tumour at least 6 months ago to take part. We are inviting both parents/caregivers of the same child to take part, but you can also take part without the child's other parent, this is completely up to you.

We are not asking parents of children who ended treatment more than five years ago to take part in the current study. Parents who find it difficult to understand written English would not be suitable for the current study. We are also not approaching parents of a child who is receiving palliative care.

Do I have to take part?

No, it is up to you to decide if you would like to take part. Whether you do decide to take part or not will have no effect on the care that your child is receiving. If you are not sure, we can try to answer any questions you may have before you decide. If you contact the researcher directly then this will act as your consent to discuss the research further. If you agree to take part you will be asked to sign a consent form.

What would taking part involve?

There are different ways in which you can express your interest in taking part. If approached at XXXX hospital, you will have the opportunity to ask questions here and can agree to participate at this point. You can also show your interest in the study by emailing the lead researcher or

completing a consent to share contact details form. Ideally, we recommend taking 48 hours to think about whether you want to take part, but this is up to you. After this, you will be asked to sign a consent form. You will then be given a questionnaire pack that will contain five questionnaires and will ask you about some of your thoughts and feelings in relation to your child's diagnosis. In total these should take about 20 minutes to complete and can be done in the clinic or at home. After you have completed them please hand them to the XXXX reception or XXXX Alternatively, if you are taking them home please send them back in the stamped addressed envelope provided. You do not need to show anyone in the clinical team your questionnaires.

Alternatively, the questionnaires have been put online and can be completed this way. The team can give you a piece of paper with the link on it, if you would prefer to complete it in this way.

What are the disadvantages of taking part?

It is not envisaged that there are any risks to you in taking part. However, we acknowledge that you may become upset when completing the questionnaires, as they include sensitive questions about some of your feelings in relation to your child's diagnosis. You do not have to answer all the questions if you do not want to and if you do become upset it is important to remember that you can withdraw from the study at any time. If you do experience any distress, then there is information in the debrief form about who you can contact.

There is one questionnaire that looks at post-traumatic stress reactions in parents. If you score highly on this, we will send you a letter informing you of this and signpost you to where you could access more help.

What are the possible benefits of taking part?

This study may not benefit you directly, but it is hoped that the information gathered in this study will help to increase our understanding about what factors may impact post-traumatic stress symptoms in parents. You will also have the option of receiving a short summary of the findings.

What if there is a problem?

If you have any concerns about any aspects of this study, you should speak to the researcher directly who will try to answer your questions. If you remain unhappy you can make a formal complaint and can contact the research supervisor, Kiki Mastroyannopoulou (K.Mastroyannopoulou@uea.ac.uk), or the Deputy Programme Director, Professor Sian Coker, (S.Coker@uea.ac.uk). If you would like to speak to someone for independent advice about participating in research in general, then you can contact the Patient Advice and Liaison Service on 01223 216 756 or pals@XXXX.nhs.uk.

What happens if I don't want to carry on with the study?

You are free to withdraw from the study without giving a reason. If you decide that you would like to withdraw this needs to be done by contacting the researcher on the details below before data analysis takes place. Up until this point if you withdraw from the study, any information that you have provided will be destroyed.

Will information be kept confidentially?

All information that is collected will be kept confidential. Relevant sections of the data collected during the study may be looked at by individuals from regulatory authorities or from the NHS Trust. All documentation including identifiable data will be kept securely. When your data is entered onto the computer your name will be replaced with a number.

The University of East Anglia is the sponsor for this study based in the United Kingdom. We will be using information from you in order to undertake this study and will act as the data controller for this study. This means that we are responsible for looking after your information and using it properly. The University of East Anglia will keep identifiable information about you for 10 years after the study has finished.

Your rights to access, change or move your information are limited, as we need to manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the study, we will keep the information about you that we have already obtained. To safeguard your rights, we will use the minimum personally-identifiable information possible.

You can find out more about how we use your information at https://portal.uea.ac.uk/information-services/strategy-planning-and-compliance/regulations-and-policies/information-regulations-and-policies/data-protection

XXXX Hospital will use your name, and contact details to contact you about the research study, and make sure that relevant information about the study is recorded for your care, and to oversee the quality of the study. Individuals from the University of East Anglia and regulatory organisations may look at your medical and research records to check the accuracy of the research study. XXXX Hospital will pass these details to the University of East Anglia along with the information collected from you. The only people in the University of East Anglia who will have access to information that identifies you will be people who need to contact you about the study or audit the data collection process. The people who analyse the information will not be able to identify you and will not be able to find out your name or contact details. XXXX will keep identifiable information about you from this study for 10 years after the study has finished.

Who has reviewed the study?

All research in the NHS and at the University of East Anglia is looked at by an independent group of people called a Research Ethics Committee, to protect your interests. This study has been reviewed and given a favourable opinion by the Social Care Research Ethics Committee.

What will happen to the results of the study?

We intend to publish the results of this study in a journal. There will be no personal details used in this.

Who is organising and funding this study?

This study is being organised by Miss Briony Westgate (Trainee Clinical Psychologist) and forms part of Briony Westgate's doctoral thesis, under the supervision of Kiki Mastroyannopoulou. The University of East Anglia are funding the study.

Further information and contact details

If you have any questions I can be contacted using the following details:

Briony Westgate
Norwich Medical School
Postgraduate Research Office
Elizabeth Fry Building
University of East Anglia
Norwich Research Park, Norwich
NR4 7TJ
Email: B.Westgate@uea.ac.uk

Appendix F

Information sheet for online sample (final version)

Examining the wellbeing of parents of children who have been diagnosed with a brain tumour

My name is Briony Westgate, I am a Trainee Clinical Psychologist and I would like to invite you to take part in a study that is looking at parents' wellbeing after their child has been diagnosed with a brain tumour. You do not have to make any immediate decisions about taking part in this online study.

What is the purpose of the project?

This project aims to looks at parents' wellbeing following their child's brain tumour diagnosis. The study will look at parents' post-traumatic stress symptoms and look at what factors might contribute to this. Post-traumatic stress symptoms can include having nightmares, trouble concentrating, and feeling numb, amongst other symptoms. There is little research into what factors might impact this in parents of children with a brain tumour and this research aims to increase our understanding of what may contribute to this.

Can I take part?

We are asking parents of children aged 4-16 years who are receiving care from a paediatric oncology service and have been diagnosed with a brain tumour at least 6 months ago to take part. We are inviting both parents of the same child to take part, but you can also take part without the child's other parent, this is completely up to you.

We are not asking parents of children who ended treatment more than five years ago to take part in the current study. Parents who find it difficult to understand written English would not be suitable for the current study. We are also not approaching parents of a child who is receiving palliative care.

Do I have to take part?

No, it is up to you to decide if you would like to take part.

What would taking part involve?

After you have provided your consent on the following page you will be presented with five questionnaires that can be completed on this online survey. The questionnaires will ask you about some of your thoughts and feelings in relation to your child's diagnosis. In total these should take about 20 minutes to complete.

What are the disadvantages of taking part?

It is not envisaged that there are any risks to you in taking part. However, we acknowledge that you may become upset when completing the questionnaires as they include sensitive questions about some of your feelings in relation to your child's diagnosis. You do not have to answer all the questions if you do not want to and if you do become upset it is important to remember that you can stop taking part at any time. If you do experience any distress, there is information at the bottom of this information page about who you can contact (the same information will also be displayed at the end of the study).

What are the possible benefits of taking part?

This study may not benefit you directly, but it is hoped that the information gathered in this study will help to increase our understanding about what factors may impact post-traumatic stress symptoms in parents.

What if there is a problem?

If you have any concerns about any aspects of this study, you should speak to the researcher directly who will try to answer your questions. If you remain unhappy you can make a formal complaint and can contact the research supervisor, Kiki Mastroyannopoulou (K.Mastroyannopoulou@uea.ac.uk) or the Deputy Programme Director, Professor Sian Coker, (S.Coker@uea.ac.uk).

What happens if I don't want to carry on with the study?

You are free to withdraw from the study by clicking off this internet page. As the study is anonymous, once you have submitted your answers you will be unable to withdraw your data.

Will information be kept confidentially?

All information you complete will remain confidential and yours and your child's names or personal information will not be collected.

The University of East Anglia is the sponsor for this study based in the United Kingdom. We will be using information from you in order to undertake this study and will act as the data controller for this study. This means that we are responsible for looking after your information and using it properly. The University of East Anglia will keep identifiable information about you for 10 years after the study has finished.

Your rights to access, change or move your information are limited, as we need to manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the study, we will keep the information about you that we have already obtained. To safeguard your rights, we will use the minimum personally-identifiable information possible.

You can find out more about how we use your information at https://portal.uea.ac.uk/information-services/strategy-planning-and-compliance/regulations-and-policies/information-regulations-and-policies/data-protection

Who has reviewed the study?

All research in the NHS and at the University of East Anglia is looked at by an independent group of people called a Research Ethics Committee, to protect your interests. This study has been reviewed and given a favourable opinion by the Social Care Research Ethics Committee.

What will happen to the results of the study?

We intend to publish the results of this study in a journal. There will be no personal details used in this.

Who is organising and funding this study?

This study is being organised by Miss Briony Westgate (Trainee Clinical Psychologist) and forms part of Briony Westgate's doctoral thesis, under the supervision of Kiki Mastroyannopoulou. The University of East Anglia are funding the study.

Further information and contact details

If you have any questions, I can be contacted on the following email address:

Email: B.Westgate@uea.ac.uk

Support you can access

If you become upset when completing this study and would like some more support, please go to see your General Practitioner (GP). You may also contact Samaritans on 116123; this service is free to call and open 24 hours a day.

There is also information about support available from the website below if you would like to read about how you can access more information and support in relation to your child's brain tumour:

https://www.thebraintumourcharity.org/get-support/

In addition, there are resources and stories on the website below from a charity that supports survivors of brain tumours, and their families:

https://successcharity.org/community/

Version 4 September 2018

Appendix G

Consent form (final version)

Briony Westgate
Norwich medical school
Postgraduate Research Office 2.30
Elizabeth Fry Building
University of East Anglia
Norwich Research Park Norwich
NR4 7TJ

Email: b.westgate@uea.ac.uk

Participant Identification Number:

University of East Anglia

CONSENT FORM

Title of Project: Examining the wellbeing of parents of children with a brain tumour

Name of Researcher: Briony Westgate (Trainee Clinical Psychologist)

Please initial box

1. I confirm that I have read the information sheet dated September 2018 (version 4) for the above study. ☐
2. I have had the opportunity to think about the information, ask questions and have had these answered. ☐
3. I understand that relevant sections of the data collected during the study, may be looked at by individuals from the UEA, regulatory authorities or from the NHS Trust. I give permission for these individuals to have access to this information. ☐
4. I understand that I do not have to take part in the study and that I am free to withdraw up to the point of data analysis without giving any reason and without my child's care being affected. ☐
5. I agree to take part in the above study. ☐

Name of Participant	Date	Signature

Please provide your home address below in case we need to contact you regarding the study:

Please provide an email address if you are happy to be contacted via email to remind you of the study (if you have consented but not sent back the questionnaires):

……

Would you like to receive a short summary of the findings via email? (please circle): **Yes/No**

If yes please provide your email address if you have not done so above:

……

Appendix H

Consent questions online (final version)

Consent questions for the online survey for parents recruited from online advertisements

1. I confirm that I am a parent of a child aged 4-16 years who has been diagnosed with a brain tumour*
2. I confirm that my child is at least 6 months post-diagnosis and has not ended treatment more than five years ago
3. I confirm that I currently live in the UK
4. I confirm that I have read the information provided for the above study
5. I understand that I do not have to take part in the study and I can exit from the study at any time
6. I agree to take part in the above study.

Appendix I

Poster (final version)

University of East Anglia

Examining the wellbeing of parents of children who have been diagnosed with a brain tumour

Researcher: Briony Westgate (Trainee Clinical Psychologist)

Research Supervisor: Kiki Mastroyannopoulou (Clinical Psychologist)

Aims

The study aims to find out more about parents' wellbeing after their child has been diagnosed with a brain tumour and look at what factors might contribute to this. There is little research into what factors might impact post-traumatic stress reactions in parents of children with a brain tumour and this research aims to increase our understanding of what may contribute to this.

Who we are inviting to participate?

We are inviting parents of children (4-16 years) who have been diagnosed with a brain tumour at least 6 months ago.

Parents who find it difficult to understand written English would not be suitable for the current study. The research team are also not asking parents of a child who ended treatment more than five years ago to take part, or a parent of a child who is receiving palliative care.

What would you need to do?

The study is a questionnaire study that will take about 20 minutes to complete. There are five questionnaires that can be done in at clinic, at home, or online. These questionnaires ask for information about some of your thoughts and feelings in relation to your child's diagnosis. They will also ask for some demographical information about you and your child.

If you would like to learn more about the study or are interested in taking part please email me on **b.westgate@uea.ac.uk**

Post-traumatic stress study: b.westgate@uea.ac.uk	Post-traumatic stress study: b.westgate@uea.ac.uk	Post-traumatic stress study: b.westgate@uea.ac.uk	Post-traumatic stress study: b.westgate@uea.ac.uk	Post-traumatic stress study: b.westgate@uea.ac.uk	Post-traumatic stress study: b.westgate@uea.ac.ukuk	Post-traumatic stress study: b.westgate@uea.ac.uk	Post-traumatic stress study: b.westgate@uea.ac.uk	Post-traumatic stress study: b.westgate@uea.ac.uk	Post-traumatic stress study: b.westgate@uea.ac.uk

Appendix J

Study advertisement (final version)

Briony Westgate is inviting parents to take part in a study that is investigating the wellbeing of parents of children who have been diagnosed with a brain tumour. The anonymous online survey will take approximately 20 minutes to complete.

The researchers are looking for parents who meet the following criteria:

- Parents (above the age of 16), living in the UK, of children who have been diagnosed with a brain tumour
- The child should be between 4 years and 16 years at the time of completion
- The child should be at least 6 months post-diagnosis
- The child should be receiving care from a paediatric oncology service

We are not asking parents of children who ended treatment more than five years ago to take part. Parents who find it difficult to understand written English would not be suitable for the current study due to the questionnaires being used. The research team are also not asking parents of a child who is receiving palliative care to take part.

If you would like more information about the study, please contact Briony on b.westgate@uea.ac.uk.

The study link is copied below:

https://uea.onlinesurveys.ac.uk/post-traumatic-stress-in-parents-online-link

Appendix K

Letter of HRA approval

Health Research Authority

Miss Briony Westgate

University of East Anglia Email: hra.approval@nhs.net
Department of Clinical Psychology, Norwich Medical School

Norwich, Norfolk

NR4 7TJ

12 March 2018

Dear Miss Westgate

Letter of HRA Approval

Study title:	**Factors that predict post-traumatic stress symptoms in parents of children with a brain tumour**
IRAS project ID:	**230003**
REC reference:	**18/IEC08/0002**
Sponsor	**University of East Anglia**

I am pleased to confirm that **HRA Approval** has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further from the HRA.

How should I continue to work with participating NHS organisations in England?
You should now provide a copy of this letter to all participating NHS organisations in England, as well as any documentation that has been updated as a result of the assessment.

Following the arranging of capacity and capability, participating NHS organisations should **formally confirm** their capacity and capability to undertake the study. How this will be confirmed is detailed in the "*summary of HRA assessment*" section towards the end of this letter.

You should provide, if you have not already done so, detailed instructions to each organisation as to how you will notify them that research activities may commence at site

following their confirmation of capacity and capability (e.g. provision by you of a 'green light' email, formal notification following a site initiation visit, activities may commence immediately following confirmation by participating organisation, etc.).

It is important that you involve both the research management function (e.g. R&D office) supporting each organisation and the local research team (where there is one) in setting up your study. Contact details of the research management function for each organisation can be accessed here.

How should I work with participating NHS/HSC organisations in Northern Ireland, Scotland and Wales?

HRA Approval does not apply to NHS/HSC organisations within the devolved administrations of Northern Ireland, Scotland and Wales.

If you indicated in your IRAS form that you do have participating organisations in one or more devolved administration, the HRA has sent the final document set and the study wide governance report (including this letter) to the coordinating centre of each participating nation. You should work with the relevant national coordinating functions to ensure any nation specific checks are complete, and with each site so that they are able to give management permission for the study to begin.

Please see IRAS Help for information on working with Northern Ireland, Scotland and Wales.

How should I work with participating non-NHS organisations?

HRA Approval does not apply to non-NHS organisations. You should work with your non-NHS organisations to obtain local agreement in accordance with their procedures.

What are my notification responsibilities during the study?

The document "*After Ethical Review – guidance for sponsors and investigators*", issued with your REC favourable opinion, gives detailed guidance on reporting expectations for studies, including: Registration of research

- Notifying amendments
- Notifying the end of the study

The HRA website also provides guidance on these topics, and is updated in the light of changes in reporting expectations or procedures.

I am a participating NHS organisation in England. What should I do once I receive this letter? You should work with the applicant and sponsor to complete any outstanding arrangements so you are able to confirm capacity and capability in line with the information provided in this letter.

The sponsor contact for this application is as follows:

Name: Sarah Green
Tel: 01603 591721
Email: sarah.green@uea.ac.uk

Who should I contact for further information?
Please do not hesitate to contact me for assistance with this application. My contact details are below.

Your IRAS project ID is **230003**. Please quote this on all correspondence.

Isobel Lyle | Senior Assessor
Health Research Authority
HRA, Room 1, Jarrow Business Centre, Rolling Mill Rd, Jarrow, NE32 3D
T: 0207 972 2496
Hra.approval@nhs.net or Isobel.lyle@nhs.net
www.hra.nhs.uk

Sign up to receive our newsletter HRA Latest

Copy to: *Ms Sarah Green, Sponsor contact, University of East Anglia R&D Dept*

List of Documents

The final document set assessed and approved by HRA Approval is listed below.

Document	*Version*	*Date*
Copies of advertisement materials for research participants [Advertisement online]	2	23 February 2018
Copies of advertisement materials for research participants [Poster]	2	23 February 2018
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Sponsor insurance]	1	12 January 2018
GP/consultant information sheets or letters [PTSS letter to parent]	1	08 August 2017
HRA Schedule of Events [HRA assessed]	1.0	23 January 2018
HRA Statement of Activities [HRA assessed]	2.0	29 January 2018
IRAS Application Form [IRAS_Form_12012018]		12 January 2018

Letter from sponsor [Sponsor letter]	1	12 January 2018
Letters of invitation to participant [Study invitation]	1	08 August 2017
Non-validated questionnaire [Demographic questionnaire]	2	23 February 2018
Other [Proposal feedback]	1	11 July 2017
Other [Response to feedback]	1	08 August 2017
Other [Cover letter for changes]	1	23 February 2018
Participant consent form [Consent to share contact details]	2	23 February 2018
Participant consent form [Consent form XXXX]	2	23 February 2018
Participant consent form [Consent form online]	2	23 February 2018
Participant information sheet (PIS) [Debrief form (XXXX)]	1	08 August 2017
Participant information sheet (PIS) [Debrief form (Online)]	1	08 August 2017
Participant information sheet (PIS) [PIS]	2	23 February 2018
Participant information sheet (PIS) [PIS online]	2	23 February 2018
Research protocol or project proposal [Study protocol]	2	23 February 2018
Summary CV for Chief Investigator (CI) [Chief Investigator CV]	1	23 October 2017
Summary CV for student [CI/Student CV]	1	23 October 2017
Summary CV for supervisor (student research) [Secondary supervisor CV]	1	06 October 2017
Summary CV for supervisor (student research) [Primary supervisor CV]	1	05 October 2017
Summary, synopsis or diagram (flowchart) of protocol in non technical language [Recruitment flowchart]	1	02 October 2017
Summary, synopsis or diagram (flowchart) of protocol in non technical language [Lay summary]	1	08 August 2017
Validated questionnaire [Impact of events scale]	1.0	26 September 2017
Validated questionnaire [Behaviour Questionnaire]	1	12 January 2018
Validated questionnaire [Social support scale]	2	23 February 2018
Validated questionnaire [Coping inventory]	2	23 February 2018

Validated questionnaire [PTSS measure]	2	23 February 2018
Validated questionnaire [ODD scale]	2	23 February 2018
18-IEC08-0002 230003 Favourable_opinion_on_further_information		12 March 2018

Summary of HRA assessment

The following information provides assurance to you, the sponsor and the NHS in England that the study, as assessed for HRA Approval, is compliant with relevant standards. It also provides information and clarification, where appropriate, to participating NHS organisations in England to assist in assessing, arranging and confirming capacity and capability.

HRA assessment criteria

Section	HRA Assessment Criteria	Compliant with Standards?	Comments
1.1	IRAS application completed correctly	Yes	No comments
2.1	Participant information/consent documents and consent process	Yes	No comments
3.1	Protocol assessment	Yes	No comments
4.1	Allocation of responsibilities and rights are agreed and documented	Yes	A statement of activities will act as agreement of an NHS organisation to participate. The Sponsor is not requesting and does not expect any other site agreement.
4.2	Insurance/indemnity arrangements assessed	Yes	*Where applicable, independent contractors (e.g. General Practitioners) should ensure that the professional indemnity provided by their medical*

			defence organisation covers the activities expected of them for this research study
4.3	Financial arrangements assessed	Yes	No funding application is being and no funding is being provided to NHS organisations in England. The Applicant will provide stationery for mail-out (refer *Statement of Activities*)
5.1	Compliance with the Data Protection Act and data security issues assessed	Yes	No comments
5.2	CTIMPS – Arrangements for compliance with the Clinical	Not Applicable	No comments
Section	**HRA Assessment Criteria**	**Compliant with Standards?**	**Comments**
	Trials Regulations assessed		
5.3	Compliance with any applicable laws or regulations	Yes	No comments
6.1	NHS Research Ethics Committee	Yes	No comments
6.2	CTIMPS – Clinical Trials Authorisation (CTA) letter received	Not Applicable	No comments
6.3	Devices – MHRA notice of no objection received	Not Applicable	No comments
6.4	Other regulatory approvals and authorisations received	Not Applicable	No comments

Participating NHS Organisations in England

This provides detail on the types of participating NHS organisations in the study and a statement as to whether the activities at all organisations are the same or different.
This is an educational study taking place at a single NHS site, therefore, there is only one site type. The Chief Investigator or sponsor should share relevant study documents with participating NHS organisations in England in order to put arrangements in place to deliver the study. The documents should be sent to both the local study team, where applicable, and the office providing the research management function at the participating organisation. If Chief Investigators, sponsors or Principal Investigators are asked to complete site level forms for participating NHS organisations in England which are not provided in IRAS or on the HRA website, the Chief Investigator, sponsor or Principal Investigator should notify the HRA immediately at hra.approval@nhs.net. The HRA will work with these organisations to achieve a consistent approach to information provision.

Confirmation of Capacity and Capability

This describes whether formal confirmation of capacity and capability is expected from participating NHS organisations in England.
The participating NHS organisations in England **will be expected to formally confirm their capacity and capability** to host this research. • The sponsor should ensure that participating NHS organisations are provided with a copy of this letter and all relevant study documentation, and work jointly with NHS organisations to arrange capacity and capability whilst the HRA assessment is ongoing. • Further detail on how capacity and capability will be confirmed by participating NHS organisations, following issue of the Letter of HRA Approval, is provided in the *Participating NHS Organisations* and *Allocation of responsibilities and rights are agreed and documented (4.1 of HRA assessment criteria)* sections of this appendix. • The Assessing, Arranging, and Confirming document on the HRA website provides further information for the sponsor and NHS organisations on assessing, arranging and confirming capacity and capability.

Principal Investigator Suitability

This confirms whether the sponsor's position on whether a PI, LC or neither should be in place is correct for each type of participating NHS organisation in England, and the minimum expectations for education, training and experience that PIs should meet (where applicable).

The Sponsor has advised that local collaboration is required at the supporting site and this has been arranged (please refer to the Statement of Activities). The Student is the CI with appropriate academic supervision and will act as PI at the site. GCP training is not a generic training expectation, in line with the HRA/MHRA statement on training expectations.

HR Good Practice Resource Pack Expectations

This confirms the HR Good Practice Resource Pack expectations for the study and the pre-engagement checks that should and should not be undertaken.
Where arrangements are not already in place, an NHS to NHS confirmation of pre-engagement checks letter would be expected research staff undertaking any of the research activities listed in A18 or A19 of the IRAS form would be expected based on standard DBS checks and occupational health clearance The Chief Investigator is an NHS employee but not employed by the participating NHS organisation

Other Information to Aid Study Set-up

This details any other information that may be helpful to sponsors and participating NHS organisations in England in study set-up.
☐ The applicant has indicated that they do not intend to apply for inclusion on the NIHR CRN Portfolio.

Appendix L

REC approval

Health Research Authority

Social Care REC
Ground Floor
Skipton House
80 London Road
London
SE1 6LH

Telephone: 0207 972 2568
Fax:

Please note: This is the favourable opinion of the REC only and does not allow you to start your study at NHS sites in England until you receive HRA Approval

12 March 2018

Miss Briony Westgate

University of East Anglia

Department of Clinical Psychology, Norwich Medical School

Norwich, Norfolk

NR4 7TJ

Dear Miss Westgate

Study title:	**Factors that predict post-traumatic stress symptoms in parents of children with a brain tumour**
REC reference:	**18/IEC08/0002**
IRAS project ID:	**230003**

Thank you for your letter of 26 February 2018, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details. Publication will be no earlier than three months from the date of this opinion letter. Should you wish to provide a substitute contact point, require further information, or wish to make a request to postpone publication, please contact hra.studyregistration@nhs.net outlining the reasons for your request.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Conditions of the favourable opinion

The REC favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements. Each NHS organisation must confirm through the signing of agreements and/or other documents that it has given permission for the research to proceed (except where explicitly specified otherwise).

Guidance on applying for NHS permission for research is available in the Integrated Research Application System, www.hra.nhs.uk or at http://www.rdforum.nhs.uk.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"),

guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of management permissions from host organisations

Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database within 6 weeks of recruitment of the first participant (for medical device studies, within the timeline determined by the current registration and publication trees).

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g. when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non-clinical trials this is not currently mandatory.

If a sponsor wishes to request a deferral for study registration within the required timeframe, they should contact hra.studyregistration@nhs.net. The expectation is that all clinical trials will be registered, however, in exceptional circumstances non registration may be permissible with prior agreement from the HRA. Guidance on where to register is provided on the HRA website.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Ethical review of research sites

NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

Document	*Version*	*Date*

Copies of advertisement materials for research participants [Advertisement online]	2	23 February 2018
Copies of advertisement materials for research participants [Poster]	2	23 February 2018
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Sponsor insurance]	1	12 January 2018
GP/consultant information sheets or letters [PTSS letter to parent]	1	08 August 2017
IRAS Application Form [IRAS_Form_12012018]		12 January 2018
IRAS Application Form XML file [IRAS_Form_12012018]		12 January 2018
Letter from sponsor [Sponsor letter]	1	12 January 2018
Letters of invitation to participant [Study invitation]	1	08 August 2017
Non-validated questionnaire [Demographic questionnaire]	2	23 February 2018
Other [Proposal feedback]	1	11 July 2017
Other [Response to feedback]	1	08 August 2017
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Summary CV for supervisor (student research) [Secondary supervisor CV]	1	06 October 2017
Summary CV for supervisor (student research) [Primary supervisor CV]	1	05 October 2017

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Validated questionnaire [Behaviour Questionnaire]	1	12 January 2018
Validated questionnaire [Social support scale]	2	23 February 2018
Validated questionnaire [Coping inventory]	2	23 February 2018
Validated questionnaire [PTSS measure]	2	23 February 2018
Validated questionnaire [ODD scale]	2	23 February 2018

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Reporting requirements

The attached document *"After ethical review – guidance for researchers"* gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The HRA website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the

service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website:

http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/

HRA Training

We are pleased to welcome researchers and R&D staff at our training days – see details at http://www.hra.nhs.uk/hra-training/

18/IEC08/0002 **Please quote this number on all correspondence** With the Committee's best wishes for the success of this project.

Yours sincerely

Dr Martin Stevens Chair

Email:nrescommittee.social-care@nhs.net

Enclosures: "After ethical review – guidance for researchers"

Copy to: *Ms Sarah Green*

Professor Ed Bullmore, XXXX

Appendix M
Capability and capacity letters

IRAS ID: 230003
Factors that predict post-traumatic stress symptoms in parents of children with a brain tumour
REC Ref: 18/1EC08/0002

Thank you for sending details of the above named study.

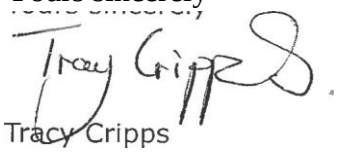
The R&D department has received the HRA Approval letter and reviewed the study documents. The project has been allocated the internal R&D reference number of A094768 Please quote this in all future correspondence regarding this study.

Capacity and capability to conduct this study at XXX is confirmed. Recruitment can commence at this site from the date of this

We would like to take this opportunity to remind you of your responsibilities under the terms of the Research Governance Framework for Researchers, Chief Investigators, Principal Investigators and Research Sponsors and to also of the requirement to notify R&D of any amendments or changes made to this study,

You will be aware that the Trust is subject to national reporting requirements for first patient recruitment within 70 days. Further details on this can be found on the NIHR website:
http://www.nihr.ac.uk/research-and-impact/nhs-research-performance/performance-ininitiating-and-delivering-research/
If you have any questions or concerns about this, please contact me.

I wish you every success with this study.
Yours sincerely



Research Governance Manager

Miss Briony Westgate
University of East Anglia
Department of Clinical Psychology
Norwich Medical School
Norwich
NR4 7TJ

18/12/2018

Dear Miss Westgate,

Confirmation of Capacity and Capability

RE: 230003 (152-11-18)

Study Title: Factors that predict post-traumatic stress symptoms in parents of children with a brain tumour

This letter confirms that XXXX has the capacity and capability to deliver the above referenced study as a PIC site. Please find attached our signed Statement of Activities as confirmation.

We agree to start this study on a date when the sponsor gives the green light to begin.

If you wish to discuss further, please do not hesitate to contact me.

Kind regards

Dr Sally Burtles
Senior Research Operations Manager

Cc. 'Kiki Mastroyannopoulou ,Sarah Ruthven

Appendix N
Letter of access

Dear Briony,
Letter of access for research — A094768

As an existing NHS employee you do not require an additional honorary research contract with this NHS organisation. We are satisfied that the research activities that you will undertake in this NHS organisation are commensurate with the activities you undertake for your employer. Your employer is fully responsible for ensuring such checks as are necessary have been carried out. Your employer has confirmed in writing to this NHS organisation that the necessary pre-engagement check are in place in accordance with the role you plan to carry out in this organisation. This letter confirms your right of access to conduct research through XXXX for the purpose and on the terms and conditions set out below. This right of access commences on 28th March 2018 and ends on 30th September 2019 unless terminated earlier in accordance with the clauses below.

You have a right of access to conduct such research as confirmed in writing in the letter of permission for research from this NHS organisation. Please note that you cannot start the research until the Principal' Investigator for the research project has received a letter from us giving permission to conduct the project and you have provided the Trust's R&D department with written evidence that you have completed GCP training from an EU institution before you start your research.

The information supplied about your role in research at XXXX has been reviewed and you do not require an honorary research contract with this NHS organisation. We are satisfied that such pre-engagement checks as we consider necessary have been carried out.

You are considered to be a legal visitor to XXXX Foundation Trust premises. You are not entitled to any form of payment or access to other benefits provided by this NHS organisation to employees and this letter does not give rise to any other relationship between you and this NHS organisation, in particular that of an employee.

While undertaking research through XXXX, you will remain accountable to your place of work, XXXX but you are required to follow the reasonable instructions of Dr Angela Kirby and Dr Hugo Ford in this NHS organisation or those given on his behalf in relation to the terms of this right of access.

Where any third party claim is made, whether or not legal proceedings are issued, arising out of or in connection with your right of access, you are required to cooperate fully with any investigation by this NHS organisation in connection with any such claim and to give all such assistance as may reasonably be required regarding the conduct of any legal proceedings.

You must act in accordance with XXXX Trust policies and procedures, which are available to you upon request, and the Research Governance Framework.

You are required to co-operate with XXXX Trust in discharging its duties under the Health and Safety at Work etc Act 1974 and other health and safety legislation and to take reasonable care for the health and safety of yourself and others while on XXXX premises. You must observe the same standards of care and propriety in dealing with patients, staff, visitors, equipment and premises as is expected of any other contract holder and you must act appropriately, responsibly and professionally at all times.

If you have a health condition or disability which may affect your research role and which might require reasonable special adjustments to your role, if you have not already done so, you must notify your employer and the Trust's R&D HR Office prior to commencing your research role at the Trust.

You are required to ensure that all information regarding patients or staff remains secure and strictly confidential at all times. Personal identifiable data must be carried securely at all times and mobile devices must be encrypted. You must ensure that you understand and comply with the requirements of the NHS Confidentiality Code of Practice (https://www.gov.uk/government/publications/confidentiality-nhs-code-ofpractice) and the Data Protection Act 1998. Furthermore you should be aware that under the Act, unauthorised disclosure of information is an offence and such disclosures may lead to prosecution. Data controllers could also be fined for a breach of the Data Protection Act 1998. You must familiarise yourself with the Trust's Information Governance Code of Conduct.

You must keep confidential any information regarding the design, conduct or management or results of any research unless authorised in writing by the Trust to disclose it. You must acknowledge the Trust's contribution in any publication arising out of this Agreement.

Subject to any agreement with your employer to the contrary (e.g. as part of a multicentre study), any Intellectual Property (IP) resulting from research carried out under this Agreement will be the property of the Trust and you will do all things necessary or desirable to give effect to the assignment of this IP.

You should ensure that, where you are issued with an identity or security card, a bleep number, email or library account, keys or protective clothing, these are returned upon termination of this arrangement. Please also ensure that while on the premises you wear your ID badge at all times, or are able to prove your identity if

challenged. Please note that this NHS organisation accepts no responsibility for damage to or loss of personal property.

We may terminate your right to attend at any time either by giving seven days' written notice to you or immediately without any notice if you are in breach of any of the terms or conditions described in this letter or if you commit any act that we reasonably consider to amount to serious misconduct or to be disruptive and/or prejudicial to the interests and/or business of this NHS organisation or if you are convicted of any criminal offence. You must not undertake regulated activity if you are barred from such work. If you are barred from working with adults or children this letter of access is immediately terminated. Your employer will immediately withdraw you from undertaking this or any other regulated activity and you MUST stop undertaking any regulated activity immediately.

Your substantive employer is responsible for your conduct during this research project and may in the circumstances described above instigate disciplinary action against you.

XXXX will not indemnify you against any liability incurred as a result of any breach of confidentiality or breach of the Data Protection Act 1998. Any breach of the Data Protection Act 1998 may result in legal action against you and/or your substantive employer.

INDUCTION AND MANDATORY TRAINING

You are responsible for familiarising yourself with the Trust's policies and mandatory training courses such as Moving and Handling, Health and Safety, Fire Training etc and be aware of the responsibility to maintain a safe environment for patients, staff and visitors

Your host Manager will ensure that you receive a comprehensive Departmental Induction. She/he will also provide you with details of Corporate Induction, research specific induction and annual Mandatory Refresher Training.

If your letter of access is for more than 3 months, you must attend Corporate Induction. Where your letter of access is for more that 12 months, you must attend annual Mandatory Refresher Training.

If your current role or involvement in research changes, or any of the information provided in your Research Passport changes, you must inform your employer through their normal procedures. You must also inform your nominated manager in this NHS organisation.

Yours sincerely

Stephen Kelleher

R&D Manager, XXXX

Appendix O
Cronbach's alpha reliability scores

Cronbach's alpha scores

IES-R

Total questionnaire= .939

Intrusion sub-scale= .884

Avoidance sub-scale= .855

Hyperarousal sub-scale=.867

ODD scale

Total questionnaire= .918

Disengaged coping sub-scale

Total sub-scale= .802

Problem-focused disengagement= .783

Emotion-focused disengagement=.751

Multidimensional Scale of Perceived Social Support

Total questionnaire= .916

Significant other sub-scale= .963

Friends sub-scale= .944

Family sub-scale= .889