



University
of Glasgow

Sharples, Philip (2016) *Experiences of care and adjustment to change in caregivers of children with autoimmune encephalitis: an interpretative phenomenological analysis*. D Clin Psy thesis.

<http://theses.gla.ac.uk/7573/>

Copyright and moral rights for this thesis are retained by the author

A copy can be downloaded for personal non-commercial research or study

This thesis cannot be reproduced or quoted extensively from without first obtaining permission in writing from the Author

The content must not be changed in any way or sold commercially in any format or medium without the formal permission of the Author

When referring to this work, full bibliographic details including the author, title, awarding institution and date of the thesis must be given

**Experiences of care and adjustment to change in caregivers of children with
autoimmune encephalitis: An Interpretative Phenomenological Analysis**

and

Clinical Research Portfolio

Philip Sharples, BSc Honours

September 2016

Submitted in partial fulfilment of the requirements for the degree of Doctorate in Clinical
Psychology

Institute of Health and Wellbeing,
College of Medical, Veterinary and Life Science

University of Glasgow

© Philip Sharples, 2016

Table of Contents

	<i>Page</i>
Declaration of originality.....	3
Acknowledgements.....	4
 <u>Chapter 1: Systematic Review</u>	 5-41
Exploring the lived experience of caregivers following paediatric traumatic brain injury: A meta-ethnography.	
 <u>Chapter 2: Major Research Project</u>	 42-87
Experiences of care and adjustment to change in caregivers of children with autoimmune encephalitis: An Interpretative Phenomenological Analysis	
 <u>Appendices</u>	 88-136
 <u>List of tables and figures</u>	
Table 1.1: Topic areas and associated search terms	11
Figure 1.1: Flow diagram of systematic study selection process	13
Table 1.2: Quality criteria scores and methodological details of included studies	15-16
Table 1.3: Themes from included studies in the meta-ethnography	18-20
Table 2.1: Super-ordinate themes and sub-themes	56



University of Glasgow | Institute of Health & Wellbeing

Declaration of Originality Form

This form **must** be completed and signed and submitted with all assignments.

Please complete the information below (using BLOCK CAPITALS).

Name PHILIP SHARPLES

Student Number 2109024s

Course Name

Doctorate in Clinical Psychology

Assignment Number/Name

Clinical Research Portfolio

An extract from the University's Statement on Plagiarism is provided overleaf. Please read carefully THEN read and sign the declaration below.

I confirm that this assignment is my own work and that I have:

Read and understood the guidance on plagiarism in the Doctorate in Clinical Psychology Programme Handbook, including the University of Glasgow Statement on Plagiarism



Clearly referenced, in both the text and the bibliography or references, **all sources** used in the work



Fully referenced (including page numbers) and used inverted commas for **all text quoted** from books, journals, web etc. (Please check the section on referencing in the 'Guide to Writing Essays & Reports' appendix of the Graduate School Research Training Programme handbook.)



Provided the sources for all tables, figures, data etc. that are not my own work



Not made use of the work of any other student(s) past or present without acknowledgement. This includes any of my own work, that has been previously, or concurrently, submitted for assessment, either at this or any other educational institution, including school (see overleaf at 31.2)



Not sought or used the services of any professional agencies to produce this work



In addition, I understand that any false claim in respect of this work will result in disciplinary action in accordance with University regulations



DECLARATION:

I am aware of and understand the University's policy on plagiarism and I certify that this assignment is my own work, except where indicated by referencing, and that I have followed the good academic practices noted above

Signature

Date 25/07/2016

Acknowledgements

I would like to thank all of the parents who participated in my research, particularly their bravery and courage recounting some difficult and distressing experiences for the benefit of this research.

Thank you to Dr Liam Dorris, Prof. Tom McMillan, and Dr Sarah Wilson for their support, guidance and encouragement and for all the time you have invested in this project and helpful feedback. I would also like to thank Dr Liam Dorris and the NHS Greater Glasgow and Clyde staff in the Paediatric Neurosciences Department in the Royal Hospital for Children for their support and persistence with recruitment.

I would like to thank my wife, Michelle, for her unconditional support and always being there for advice, encouragement and organisation. Thank you to my boys, Aiden and Olly, for their patience and for always making me smile even through the stressful times. Finally thank you to my family, friends and fellow classmates for their tolerance and understanding over the last three years, which undoubtedly has gotten me this far.

Chapter 1: Systematic Review

Exploring the lived experience of caregivers following paediatric traumatic brain injury: A meta-ethnography

Philip Sharples

Mental Health and Wellbeing

University of Glasgow

1st Floor, Admin Building

Gartnavel Royal Hospital

1055 Great Western Road

Glasgow, G12 0XH

E-mail: p.sharples.1@research.gla.ac.uk

Word count with quotes: 7,817

Word count without quotes: 6,615

Preparation in accordance with guidelines for submission for *Developmental Neurorehabilitation* (see appendix I)

Abstract

Background: Traumatic brain injury (TBI) is the leading cause of acquired disability amongst paediatric populations. The quantitative literature indicates a reciprocal relationship between caregiver functioning and child outcomes following TBI, and an increasing emphasis on adjustment and coping in caregivers.

Aims: To systematically review the qualitative literature on the post-discharge experiences of caregivers of children with a moderate to severe TBI.

Methods: A systematic search of electronic databases, hand search of selected journals and reference sections of identified papers were conducted. Articles were appraised using Walsh and Downe's (2006) quality appraisal framework, and synthesised using a meta-ethnography approach (Noblit and Hare, 1988).

Results: Nine studies were identified for review. Three constructs emerged: psychosocial impact on caregiver and family functioning; adjustment, coping and resilience; and relationships and support. Results suggest that the impact on caregiver adjustment is underpinned by several inter-related factors. The direct consequences of the sequelae of the TBI, perceived 'loss of the previous' child, uncertainty and the effect on the family system were prevalent. The repercussions of these experiences can be further mediated by the caregiver's method of coping, and support from services, family and the community.

Conclusions and implications: The impact of paediatric TBI on caregivers and the family system should be actively considered by services and policy makers. Recommendations are made regarding options for supporting families and future research.

Keywords: Traumatic Brain Injury, Paediatric, Caregiver, Qualitative Systematic Review, Adjustment

1. Introduction

The foremost cause of acquired disability and death in children and young people is traumatic brain injury (TBI) (Keenan and Bratton, 2006). Paediatric TBI can result in enduring psycho-social outcomes for the child (Trenchard, Rust and Bunton, 2013). There is often higher prevalence of neuropsychological, behavioural, adaptive and academic deficits in the child (Fay et al, 2009). Caregivers of these children can experience increased risk of psychological problems, care-giver burden, and family dysfunction (Stancin et al, 2008; Rashid et al 2014). Conversely, family functioning can affect the psychological outcome in the injured child (Lax-Pericall and Taylor, 2014). Therefore, the child's impairments, particularly behavioural outcomes, can be predictive of poor family outcomes, and elevated parental distress can impact the child's recovery (Taylor et al, 2001). This reciprocal interaction between child outcomes and parental functioning has resulted in an increasing emphasis on including the family in rehabilitation of paediatric TBI through parenting interventions and emotional support.

A systematic review of the efficacy of parenting interventions for parents of children with a TBI found that interventions could improve parenting skill and adjustment, and reduce emotional and behavioural difficulties in the child (Brown et al 2013b). Parental interventions typically draw on behavioural and social learning theories to modify the child's behaviour. There is strong evidence for parental management training for children with perceived emotional and behavioural difficulties in the general population (Kazdin, 1997). These include the Triple P-Positive Parenting Program (Sanders, 1999) and the Incredible Years Program (Webster-Stratton, 1990). In order to apply interventions of this

kind to parents of children with a TBI it is important to identify and develop the components which are pertinent to this population. Sanders and Kirby (2012) identified the importance of consumer engagement when developing and adapting the core components of parenting interventions. Qualitative synthesis exploring the experience and knowledge of the consumer, in this case the parents, could improve the relevance and quality of parenting interventions for this population.

After TBI, families may progress through a grieving process, similar to the death of a loved one. This process can be more complex and enduring with children, often re-emerging when they reach important life transitions and developmental milestones. Lezak (1986) argues that families typically experience six stages of adjustment in response to TBI in a family member to varying degrees. While families often report positive emotions in response to their family member returning home, they later experience a period of anxiety and confusion as they begin to notice changes. This is typically followed by stages characterised by depression, guilt and despair and then by a period of mourning when the permanency of the changes become apparent. The final stage involves the family re-organising and rebuilding their lives, this can involve reinterpretation of their relationship with their loved one. Martin (1988) noted that long term care needs, changes in personality and cognition, and uncertainty over recovery have a significant impact on family adjustment to paediatric TBI. Financial difficulties and limited practical and rehabilitation support in the community can also increase family stress. Clark, Stedmon and Margison (2008) question the validity of stage models of grief and adjustment for parents of children with TBI. They argue that the prolonged grieving process and uncertainty may be explained by more flexible models such as the dual process model of grief (Stroebe and Schut, 1999). A synthesis of the qualitative research may contribute to understanding the theoretical underpinnings of adjustment in paediatric TBI.

An increasing number of qualitative studies focus on caregiver experience post-paediatric TBI. Some explore experiences at a particular stage of the child's recovery (Rosignano et al, 2013), and others focus on a specific aspect of the child's cognitive or social functioning (Ward et al, 2004; Gauvin-LePage and Lefebvre, 2010), or the severity of the TBI (Gagnon et al, 2008). There is currently no synthesis of qualitative research on caregiver experience of caring for their child, despite developments in this field. A qualitative synthesis of studies exploring the post-discharge experience of caregivers of children with a TBI could inform the development and validation of new and existing parenting interventions and outcome measures.

1.1 Aims

- To explore caregiver experiences of caring for a child with a moderate to severe TBI post-discharge from hospital.
- To critically appraise the qualitative research within this area.
- To synthesise and discuss key concepts from the qualitative literature on experiences of caregivers caring for children with a TBI.

1.2 Review question

What are caregiver's experiences of providing care to a child with a moderate to severe TBI post-discharge from hospital?

2. Method

This synthesis involved three key stages: a systematic search of key databases to identify relevant studies; quality appraisal of included studies; and meta-ethnography to identify important themes and concepts (Noblit and Hare, 1988).

2.1 Search strategy

The following electronic databases were searched: Ovid: Medline (R) (1946 – present); Embase (1947 – present); PsychINFO; PsycARTICLES; Psychology, Behavioural and Sciences Collection; and Pubmed. Due to the recognised variability in the clarity and consistency of titles and indexing of qualitative studies (Ring et al, 2011), a contents list search of two key journals (Brain Injury and Qualitative Health Research) was conducted from the past 5 years. An additional hand search was conducted to find further relevant studies in the reference sections of included papers.

2.2 Search terms

The following topics were identified as relevant: brain injury, child, parents, and qualitative methods. Key search terms were identified for each of these topic areas by referring to relevant search terms used in previously identified studies. Each topic in table 1.1 was linked using AND commands.

Table 1.1: Topic areas and associated search terms

Topic	Search Term
Brain injury	Head injury OR Traumatic Brain Injury OR Acquired Brain Injury OR Neuro\$ OR Craniocerebral Trauma OR Brain Damage OR ABI OR TBI
Child	Child\$ OR Infant\$ OR Adolescent\$ OR Paediatric
Parent	Mother OR Father OR Parent\$ OR Family OR Caregiver
Qualitative Research	Qualitative OR Interpretative Phenomenological Analysis OR Grounded Theory OR Thematic Analysis OR Content Analysis OR Focus Group\$

2.3 Inclusion criteria

- Studies utilising qualitative methodology for the collection and analysis of data.
- Studies gathering information from the primary caregiver of a child with a moderate to severe TBI.
- Studies primarily exploring caregiver's experiences of providing care for their child following discharge from hospital.
- The child with a TBI must be 0-18 years of age at the time the study was conducted.
- Studies published in English
- Studies published in peer-reviewed journals

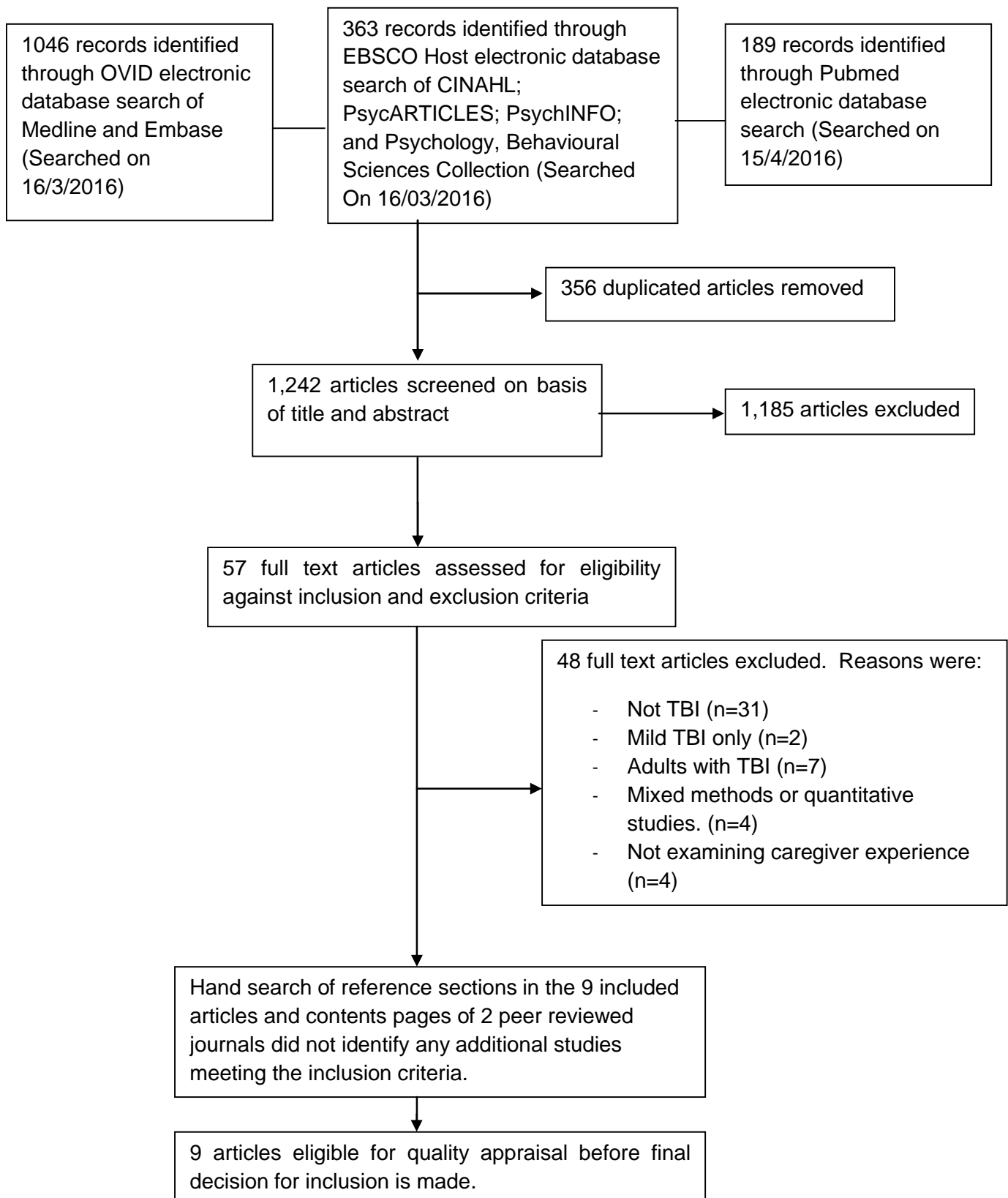
2.4 Exclusion criteria

- Studies focusing on primary caregivers of adults over the age of 18
- Studies which include children who have experienced non-traumatic neurological deficits, e.g., neurodevelopmental disorders, neurodegenerative disease, brain tumour, stroke or other acquired brain injuries.
- Studies utilising quantitative or mixed methods to collect and analyse data.
- Studies primarily focussing on the child's experiences (e.g. health related quality of life)
- Book chapters
- Non-English language studies
- Non-peer reviewed journals.

2.5 Results of systematic search

Figure 1.1 illustrates the results of the systematic search.

Figure 1.1: Flow diagram of systematic study selection process



2.6 Quality ratings

Included studies were rated against the summary framework for appraising qualitative research developed by Walsh and Downe (2006). This framework is based on subjectivist and hermeneutic epistemological stances, and was developed through critically reviewing and synthesising 'essential' criteria from other quality appraisal frameworks. The framework contains 53 items, which form the 12 essential criteria. Included studies were scored against each item and if they achieved over 50% within each individual criterion, then the criterion was satisfied (see appendix II). An independent researcher (trainee clinical psychologist) also rated a sub-sample of three included studies to improve reliability, and address any bias in the original ratings. Inter-rater reliability was high for the individual item scores (90%) and the overall criteria (97%). Unanimous agreement was reached through discussion of any discrepancies in individual scores.

When considering the inclusion of studies, balance was sought between the quality, relevancy and the importance of their findings to the research question (Edwards et al, 2000). Despite some variability in the quality ratings across studies they all contributed to the research question and therefore were all included. Concept maps and data tables were maintained when comparing and translating themes between studies. This enabled the researcher to identify concepts and interpretations from higher quality studies (see appendix IV for a worked example). Table 1.2 provides a description of the key methodological aspects of the studies and the quality ratings.

Table 1.2: Quality criteria scores and methodological details of included studies

Author (Year)	Country	Qualitative approach	Participants (Gender)	Age of child (severity of TBI)	Quality rating
Brown et al (2013a)	Australia	Inductive thematic analysis	10 caregivers (8 female, 2 male) 5 health professionals	5-17 years (mod-severe)	11/12
Bruce and Chisholm (2001)	Canada	Content analysis	10 caregivers (not specified)	5-15 years at the time of injury (mild- severe)	9/12
Clark, Stedmon and Margison (2008)	UK	Interpretive Phenomenological Analysis (IPA)	10 (all female)	0-16 years at time of interview (mod- severe)	12/12
Gauvin- Lepage, Lefebvre and Malo (2015)	Canada	Miles and Huberman (2003)	7 caregivers (6 female, 1 male), 6 adolescents & 5 rehabilitation professionals	14-17 years old at the time of study (mod- severe)	10/12
Jones, Hocking and Wright-St	New Zealand	Grounded theory	7 (4 female, 3 male)	4-11 years (severe)	12/12

Clair (2010)					
Kirk et al (2014)	UK	The framework approach	29 (20 female, 9 male)	3-16 years at time of injury (severe)	10/12
Robson, Ziviani and Spina (2005)	Australia	Thematic content analysis	6 (5 females, 1 male)	4.5-10.5 years (mod-severe)	11/12
Roscigno and Swanson (2011)	USA	Descriptive phenomenological framework	42 (34 females, 8 males)	6-18 years at time of injury (mod-severe)	11/12
Wharewera- Mika et al (2016)	New Zealand	Thematic analysis	21 (16 female, 5 male)	<2 years at time of injury (serious head injury)	10/12

2.7 Method of synthesis

Meta-ethnography (Noblit & Hare, 1988) is considered a leading method for synthesising qualitative research (Ring et al, 2011) and was used to synthesise the key concepts in the included papers. It aims to translate the concepts raised by different papers into one another. Noblit & Hare (1988) outline 7 key phases:

1. **Getting started** involves establishing a research question.

2. **Deciding what is relevant to the area of interest** involves determining the scope of the synthesis.
3. **Reading the studies** involves carefully and repeatedly reading selected studies and highlighting key concepts.
4. **Determining how the studies are related** involves identifying how the studies correspond to each other by listing the main concepts from each study and identifying recurring themes.
5. **Translating the studies into one another** involves comparing the key concepts and interactions from the different studies.
6. **Synthesising translations** involves transposing concepts and interpretations from studies into encompassing concepts, and third order interpretations.
7. **Expressing the synthesis** involves communicating the synthesis in the most appropriate format.

There are three key methods of 'synthesising translations', namely: reciprocal translational analysis, refutational synthesis, and line-of-argument synthesis. Reciprocal translational analysis is used to synthesise similar concepts across studies; refutational translations synthesise contrasting concepts; and line-of-argument synthesis aims to build an interpretation from the key concepts highlighted across studies.

3. Results

All of the papers included in this review were of good quality, with scores ranging from 9 and 12 out of 12 (see appendix III for breakdown of quality scores). All the studies were included in the synthesis, however more weight was given to the methodologically stronger papers (Brown et al, 2013a; Clark, Stedmon, and Margison, 2008; Jones,

Hocking & Wright-St Clair, 2010; Robson, Ziviani and Spina, 2005; Roscigno and Swanson, 2011), predominantly due to the richness and depth of their analyses.

The themes from the selected papers are summarised in table 1.3.

Table 1.3: Themes from included studies in the meta-ethnography

Author (Year)	Themes
Brown et al (2013a)	<ul style="list-style-type: none"> • TBI has a wide-ranging impact on a child • Parents' emotional experience is intense, overwhelming, and ongoing • Effective parenting becomes more challenging • Burden of care • Relationships with family and friends • Perceptions of connection, empowerment and support • Coping strategies employed
Bruce and Chisholm (2001)	<ul style="list-style-type: none"> • Contributing factors to the injury event • Causes of uncertainty • Factors associated with uncertainty • Management of uncertainty • Consequences of uncertainty
Clark, Stedmon and Margison (2008)	<ul style="list-style-type: none"> • Changes to and loss of the past child • Physical, psychological and social effects on mother • Mother's process of coping and support

	<ul style="list-style-type: none"> • Changed roles • Effects on the whole family • Contact with services
Gauvin-Lepage, Lefebvre and Malo (2015)	<ul style="list-style-type: none"> • Family characteristics and it's influences • Positive family strategies • Family and social support • Management of occupational aspects • Contribution from the community and health professionals
Jones, Hocking and Wright-St Clair (2010)	<ul style="list-style-type: none"> • Structuring for security • The process of holding things together • The process of joining my child with others
Kirk et al (2014)	<ul style="list-style-type: none"> • The accident and its immediate aftermath • Being on the intensive care unit • Moving to the ward • Coming home • Returning to school
Robson, Ziviani and Spina (2005)	<ul style="list-style-type: none"> • Emotional journey • Significant relationships • Services • Coping strategies
Roscigno and Swanson (2011)	<ul style="list-style-type: none"> • Grateful to still have my child • Grieving for the child I knew • Running on nerves • Grappling to get what my child and family need
Wharewera-Mika et al (2016)	<ul style="list-style-type: none"> • Impact

	<ul style="list-style-type: none"> • Support • Information
--	--

Three key constructs were identified from the meta-ethnography, which provided a line of argument synthesis: psychosocial impact on caregiver and family functioning; adjustment, coping and resilience; and relationships and support.

3.1 Psychosocial impact on caregiver and family functioning

This third order concept refers to the complex issues that affect psychological and social wellbeing in caregivers. Participants of some studies described leaving the safety of hospital to the community as particularly challenging. One study emphasised that this was the first time participants processed the consequences of the traumatic event:

‘I think when you’re in hospital it’s very sort of um... very unreal environment and still very numbing and then when you come home you sort of start to realise what’s happened and... Even though you know it was very serious when you’re in hospital, you don’t realise what extent until you get home and you’re in your own house and you’ve got time to sort of sit and think about things...’ (Robson, Ziviani and Spina, 2005, pg.49)

A further prominent theme was managing the effects of the TBI, particularly changes in their child’s behaviour and emotion regulation, which became more apparent on return home from hospital. Participants commonly described feeling distressed and exhausted in response to these changes in their child, this response was often mediated by the unpredictability and inappropriateness of their child’s behaviour:

‘You just didn’t know from one minute to the next... what was he going to be like. He could go be really sweet one minute and the next minute he could go crazy or anything. You just didn’t know what to expect.’ (Bruce and Chisholm, 2001, pg 14)

Many parents reported changes in their child’s cognitive, physical, personality and social functioning as distressing. The uncertainty parents experienced regarding how to manage their child’s behaviour, the permanency of the child’s difficulties, the cause of their behaviour and longer term outcomes resulted in persistent worry and anxiety:

‘How is she going to cope as she gets older? And I guess when they’re little, it’s still there but it’s sort of much further in the background, but then as they’re getting older and older it’s like what is she going to do when she does finish school, is she gonna be able to manage by herself, living by herself, and what kind of job is she going to have... the future is always really quite prominently there... [it’s] scary...’. (Brown et al, 2013a, pg. 1574)

A prominent concept, particularly amongst the highest quality studies (Brown et al, 2013a; Clark, Stedmon, and Margison, 2008; Jones, Hocking and Wright-St Clair, 2010; Roscigno and Swanson, 2011), was mourning the loss of the child they knew before the TBI and needing to re-establish their relationship:

‘My child, before the wreck she was active, very popular in school. She was a volleyball player... She had so many plans, you know, she had scholarships... She wanted to play Olympic volleyball... I see her now, and I look at her now, and I remember what she was like before’. (Roscigno and Swanson, 2011, pg. 1417-1418).

Caregivers responses to the loss of their ‘previous’ child were mediated by the age of the child, severity of the injury and acceptance of the permanency of the TBI sequelae. The experience of trauma was reported, predominantly by the highest quality studies (Brown

et al, 2013a; Clark, Stedmon, and Margison, 2008; Robson, Ziviani and Spina, 2005), in response to witnessing the event and/or its repercussions or the consideration that their child may not survive:

‘You never forget that, never, I thought she was dead, I went over and I couldn’t go near her, I don’t know why but I couldn’t, I was near her but I couldn’t touch her because I thought she was dead’. (Kirk et al., 2014, pg. 305)

The impact of the trauma for some parents resulted in difficulties in processing the event and a persistent emotional reaction to associated triggers. Some caregivers expressed feelings of guilt and self-blame for the circumstances around their child’s injury or anger towards other family members who they blamed:

‘I knew he needed help... but then I’d be like ‘why should I give a [expletive] about you? You did this. I hope it hurts and I’d say that, I’d get real nasty (Mother, Dale, referring to ex-partner who was involved in motor vehicle accident)’. (Brown et al, 2013a, pg. 1574)

Caregivers commonly described the impact of the TBI in terms of the relationship between the individual child, themselves and other family members. The direct impact of the TBI, caregiving demands, role changes, and differences in coping between parents were commonly described as having negative effects on the family system. This was exacerbated by ineffective communication, financial hardship, feelings of anger, resentment and guilt. It was recognised more often in the highest quality studies (Brown et al, 2013a; Clark, Stedmon, and Margison, 2008; Robson, Ziviani and Spina, 2005; Roscigno and Swanson, 2011) that siblings may experience trauma and have difficulty accessing support from their parents due to an increased focus on the injured child. Siblings could also experience difficulties in their relationship with the injured child, particularly if behavioural or emotion regulation difficulties were present:

‘Bad feeling all around, really, with everybody. The whole family. Cos, it affects everybody... it’s the same for his brother and sister and for Kevin (Stepfather)... then that affects my relationship with the two children, because then I’m having to, then, step in and stop arguments, and I think they think, that I’m being too lenient with Sean’. (Clark, Stedmon, and Margison, 2008, pg.576)

3.2 Adjustment, coping and resilience

Participants reported various ways of coping with distress in an attempt to foster resilience and adjustment to the traumatic events, changes in their child, and their uncertainty regarding the future. Caregivers perceived their child as vulnerable, and reported the need to protect their child, emotionally and physically, to reduce the likelihood of further harm. Many parents and families attempted to manage the immediate environment, organise strict routines and protocols, remain close to their child and maintain vigilance in an effort to protect them:

‘He will just not do what you want him to do. He will run away

Child: I will run out of the gate

Mother: Yes. Climb the tree is our latest thing. He knows that I get really upset when he climbs the tree

Child: Mum thought that I was going to fall down’. (Jones, Hocking, and Wright-St Clair, 2010, pg. 8)

The ability of parents to protect their child was affected by additional demands, such as work, other dependents, or being a single parent. Caregivers, predominantly in the highest quality studies (Brown et al, 2013a; Clark, Margison and Stedmon, 2008; Jones, Hocking and Wright-St Clair, 2010; Robson, Ziviani and Spina, 2005; Roscigno and

Swanson, 2011), also expressed the need to encourage recovery and independence, which could be challenging and result in ambivalence about change. Despite this most participants were motivated to facilitate recovery and rehabilitation to improve their child's outcomes, reduce their own uncertainty about the future, re-integrate the child into society, and to see signs of their 'old' child returning:

'[Because of my daughter's age] I felt like I needed to step back and let this professional work with her to teach her to be a self advocate, because that's what she needs to go forward and be successful and have a sense of confidence... her confidence has taken a real hit in this whole experience'. (Roscigno and Swanson, 2011, pg.1420)

Caregivers described various coping strategies to foster resilience to the psychosocial impact of their experiences. A key strategy reported by many parents, particularly in the highest quality studies (Brown et al, 2013a; Clark, Stedmon, and Margison, 2008; Jones, Hocking and Wright-St Clair, 2010; Robson, Ziviani and Spina, 2005; Roscigno and Swanson, 2011), was cognitive reappraisal of their situation, the changes in their child, and the future. This often involved acceptance of the child's difficulties, recognition and positive reframing of their child's recovery, and making favourable comparisons to others:

'Your world changes, your idealism of a perfect world and a perfect family, changes. And you just adapt to what's given to you and you try and make the best of what's there'.
(Brown et al, 2013a, pg. 1579)

The use of coping strategies was reportedly mediated by pre-existing coping strategies, personality and determination, spiritual beliefs, socioeconomic status and gender, with one study indicating that men found coping and adjusting more challenging. Caregivers, predominantly in the highest quality studies (Brown et al, 2013a; Clark, Stedmon, and Margison, 2008; Jones, Hocking and Wright-St Clair, 2010; Robson, Ziviani and Spina,

2005; Roscigno and Swanson, 2011) described problem focussed coping strategies, such as problem solving, researching information and services, monitoring and protecting their child, advocating for their child and involvement in planning treatment and rehabilitation:

‘When you’re in a situation like that, it’s crisis led. You’re fighting fires, so, whatever the issue is.... you’re very focussed about, ‘OK, well, what do we need to do for that’. (Clark, Stedmon, and Margison, 2008, pg. 574).

Avoidant coping strategies, such as denial, use of substances, withdrawing into work or other activities, and suppression or avoidance of difficult thoughts, emotions and associated triggers were also evident:

‘I think women are very good, and mothers are very good, at masking problems. To the point where people think ‘They’re so strong, they look after their family, and they go to work, they do this, they do that, they keep this going’. But if they really knew that you’d eaten your guts from the inside-out...that whole time...’. (Brown et al, 2013a, pg.1578)

3.3 Relationships and support

This third order construct refers to the use of social support from immediate and extended family and friends, the community, health, social, and education professionals. All studies, irrespective of quality, referred to the largely positive impact of social support from extended family, friends and the community. Family and friends who were willing to share and endure the care-giving burden, who had an understanding of the injured child’s difficulties, and who were accepting of the child were seen as particularly supportive by caregivers:

‘You did all bond and... you kind of get through it and you’re very supportive of each other..., I think family support has been really important. And not any one person in particular but from the whole thing really’. (Clark, Stedmon and Margison, 2008, pg. 575)

While social support from family and friends was largely recognised as having a positive impact on caregiver adjustment, this support was not always widely available. Some recognised the importance of their spouse in sharing the burden of care and for emotional support, particularly if there was strong communication, and validation of the primary caregivers’ role and emotions:

‘I honestly believe I couldn’t have gone through it on my own, and he has said the same thing. We just virtually leant on each other the whole time’. (Robson, Ziviani and Spina, 2005, pg. 50).

Despite the availability of family, friends and professionals, some caregivers reported that they had limited understanding of their experiences, which resulted in them feeling isolated. Many studies reported a need for caregivers to share their story, and access information and support from other people who understood, such as support groups, and other parents caring for a child with TBI:

‘There’s really not... a support group for that.. in Australia... I was... thinking I really want to... maybe set up a support group, but then... because I ended up suffering depression... even now I’s like ‘Yeah, that would be really good thing to do’, but... emotionally, I can’t do it because it just takes too much out of me’. (Brown et al, 2013a, pg. 1578)

All studies highlighted that caregivers often evaluated post-discharge support from health and social care services as variable. Many caregivers described feeling abandoned,

anxious and isolated on their return home. Participants expressed the need for guidance, information and support from professionals with regards to how to manage and understand their 'new' child and their behavioural, cognitive and emotional difficulties:

'You feel so alone and you feel, like I say, you're dealing with all this stuff that you've not got a clue really what you to do, you know what I mean... it's like have you got a manual for this child? Because I don't know who he is and I'm trying to look after him the best I can ... we'd been left with this child, with all these things going wrong with him and, as far as they were concerned, they'd sent him home... you just feel so alone and you're with this child that you don't really know what you're dealing with'. (Kirk et al., 2014, pg. 308)

In the absence of this professional support, some caregivers experienced uncertainty, which may result in the family 'protectively bonding' and becoming resistant to future professional input (Clark, Stedmon, and Margison, 2008). Some studies emphasised the ongoing struggle many families reported while attempting to obtain the required support for their child:

'She (the child) was threatening suicide and I just kept ringing people up and banging on doors until I got someone to actually listen to me... I got told about (service) so I rang them up and I said you've just got to help and they gave me an appointment and we went to see them... I felt that they were incredibly busy and they said things to me like 'we don't see her (the child) as someone who needs our help, we've got worse cases to deal with', and I was really disappointed actually'. (Wharewera-Mika et al, 2016, pg. 275)

Some participants also recognised the need for emotional support, such as counselling, for them and their family, in response to the trauma and their emotional wellbeing:

'I found that... my husband... really needed someone to talk to, and they were all supporting me... there's nothing for the men. They're treated rather badly, ignored... in hindsight I think that a lot more could've been done for the men'. (Brown et al, 2013a, pg.1575)

It was important that professionals were empathic, trustworthy, compassionate, and genuine when supporting families to meet their needs. Caregivers also expressed the need for accessible, co-ordinated and sustainable services, communicating with each other about their child.

Participants in most studies referred to the variability in education staff's understanding regarding the impact of their child's TBI on their learning and support needs. They felt this knowledge was important so the school could adapt to their child's needs, and avoid misattributing difficulties as inattentiveness or purposeful disruption. Some caregivers emphasised the role of the health service to provide training and information for education staff:

'[His teachers were always telling him], "You're faking it," or, "You're just looking for an excuse not to do something," or "You're just lazy," so, he's taken quite a bit of ridicule... [they were] going to fail him out of the fourth grade, or retain him in the fourth grade... What he did do was correct... He just can't do it as fast as they want him to do it.... I felt like it was going to be really detrimental... [to his] self-esteem, being one. It's just going to help reassure him he's a failure'. (Roscigno and Swanson, 2011, pg. 1419)

It was also recognised that the return to school would be challenging for the child and the family due to the length of time the child has been absent, cognitive sequelae limiting their capacity to learn, difficulties reintegrating with social group due to perceived changes in the child, and increased risk of bullying.

4. Discussion

Three main themes were identified from qualitative studies exploring caregivers' experiences of caring for a child with TBI. These were: psychosocial impact on caregiver and family functioning; adjustment, coping and resilience; and relationships and support. Caregivers often reported deterioration in their own and their family's wellbeing and functioning due to many chronic stressors associated with the child's injury. Caregivers and families' ability to manage these stressors differed depending on the coping strategies employed, adjustment to changes in the child, and the quality of support they received from family, friends, and professionals.

4.1 Theoretical and research implications

The identified constructs suggest that caregivers' experiences and responses to their child's TBI are underpinned by numerous and complex factors. A clear sense of grief resonated throughout the studies in response to the psychological loss of the child they once knew, however this was often interpreted differently compared to the death of a loved one. These experiences resonated across many of the studies irrespective of the length of time post-TBI. While Martin (1988) acknowledged the complexity of the grieving process in paediatric TBI, the results from this synthesis suggest that caregivers do not necessarily grieve in a linear, stage process. Some researchers describe this concept as chronic sorrow (Olshanky, 1962). This appears to be particularly relevant for caregivers of children with TBI and other neurological impairments as a sense of loss may be re-evoked as the child fails to reach developmental milestones or struggles with transitions. This prolonged experience of loss is also apparent in the literature for caregivers of children with acquired brain injuries, where mothers reported having to re-establish a relationship

with their 'different' child (Guerriere and McKeever, 1997). This synthesis suggests that the ongoing experiences of loss and adjustment may be more consistent with Stroebe and Schut's (1999) dual process model, whereby families oscillate between loss orientated and restoration orientated coping, rather than progression through discrete stages of adjustment.

Changes in their child's behaviour and emotion regulation often increased the reported stress and anxiety in caregivers and families, which is also reported in the quantitative literature (Taylor et al., 2001). While some studies describe the family becoming closer and adopting new roles, others report strain on relationships between family members. These concepts are consistent with systems theory, which propose that a significant event, such as a TBI in a child, disrupts the homeostasis, roles and patterns of interaction within the family system (Verhaeghe, Defloor, and Grypdonck, 2005). Trauma, guilt and anger towards other family members may also impede on family functioning.

The concept of protection was emphasised by many families, which could limit the child's recovery if caregivers are unable to manage their anxieties and subsequently provide their child with opportunities to develop. Clark, Stedmon and Margison (2008) suggest that the need to protect their child could be explained by attachment theory whereby proximity seeking behaviours in the child or the parent are triggered in response to the traumatic event. The trauma may also have changed the core beliefs the parent held regarding the safety of their child, which they term a 'loss of benign sense of vulnerability'. These proximity seeking, protective behaviours may also be a manifestation of problem focused coping as the parent attempts to control the environment to keep their child safe.

Family coping and resilience were key constructs emerging from this meta-synthesis. Three key coping strategies appeared to be used by caregivers: problem focused coping;

emotion focused coping; and avoidant coping. Coping Theory (Lazarus and Folkman, 1984) distinguishes between problem and emotion focused coping. While both of these strategies can be helpful, excessive use of problem focused coping in uncontrollable situations can lead to increased distress (Oddy and Herbert, 2003). Some caregivers adopted emotion focused coping strategies, recognising and reinterpreting the chronic and enduring changes in their child. Whereas other caregivers remained uncertain about the future, using a problem focused approach, such as seeking support and information from their family, peers and professionals.

4.2 Clinical and practical implications

This synthesis suggests that caregivers have unmet information, emotional, and practical support needs upon discharge from hospital. Service managers may wish to consider the provision of formal post-discharge support, involving a central, co-ordinating professional who could be the point of contact for families. This would enable the service to pre-empt many of the psychosocial consequences for families and consider appropriate interventions or resources to prevent these issues escalating. Families may benefit from reassurance and information to normalise their anxieties regarding uncertainties about the child's presentation or recovery. This co-ordinating professional could also liaise with other agencies, such as education services, to share information about the child's TBI. Effective collaboration and communication between health, education and family has also been recognised within the paediatric ABI literature (Andersson, Bellon, and Walker, 2016). Caregivers may also experience ongoing trauma in response to the circumstances surrounding the traumatic event, or from being in paediatric intensive care, which can often be overlooked by services. Health professionals may wish to consider screening primary caregivers and families for trauma, and refer them to appropriate services where necessary.

Social support was commonly reported to be a positive coping mechanism for many caregivers, but this often depends on the presence, strength and willingness of the family system to continue to provide this support. Therefore, it is important that service managers and clinicians consider each family's existing support network and how health and social care services can support the varying needs of the individual caregiver and the family system.

Individual interventions such as adapted parental skills training, psychoeducation and problem solving therapy have demonstrated effectiveness for this group and could be considered for caregivers who are unsure of how to parent their 'new child' or children who display high levels of challenging behaviour (Brown et al, 2013b). Combined acceptance and commitment therapy (ACT) and stepping stones positive parenting program have also demonstrated a reduction in behavioural and emotional difficulties for children with an ABI and improvement in parenting management strategies (Brown et al, 2014). Many of these treatments involve multiple components, therefore further research investigating the efficacy of the specific interventions is required.

Systemic interventions may also be beneficial for families who have particular difficulty adjusting to their child's TBI. Caregivers consistently expressed the need to share experiences and information with other parents of children with TBIs. The provision of support groups within local communities for families of children with neurological impairments could be a valuable resource in order to reduce anxiety, and provide training and information. Families may also benefit from adapted systemic family interventions, which could enhance communication, problem solving and knowledge within the family about TBI, facilitating role transitions and adjustment.

4.3 Limitations

There are several recognised limitations in meta-ethnography, possibly extending to this synthesis (Ring et al, 2011). The interpretation of the studies may have been skewed by the researcher's own bias or inadvertent omission of information when translating studies into third order constructs, despite efforts to reduce bias through maintenance of a reflective log and regular clinical supervision. It is possible that some studies may have been missed in the initial search due to variability in how qualitative studies are indexed (Ring et al. 2011). The effect of this was reduced by conducting hand searches through reference sections of included articles and content pages of two journals. The decision to include all articles meeting inclusion criteria, despite some variability in quality scores, may have affected the findings. Despite this the decision to exclude studies on the basis of quality is still debated in qualitative systematic reviews (Carroll, Booth and Lloyd-Jones, 2012). The decision to omit non-published dissertations, non-English language studies and book chapters from this review may have restricted the synthesis and generalisability of the themes. The generalisability of the findings could also be affected by the heterogeneity of the population, in terms of the child's age, cause of injury (non accidental versus accidental), severity (moderate to severe), and time since the injury (ranging from 1 month to over 12 years). Furthermore, all of the studies were conducted in developed countries, and the majority of the caregivers interviewed were female.

4.4 Future research

High quality qualitative research is required within this area in order to develop and refine the constructs found in this synthesis. Further research may wish to consider the specific impact of paediatric TBI on male caregivers, as they are often an overlooked and vulnerable population (Wade et al, 2010). Future research into the prevalence of trauma

within family systems and the resulting consequences on caregiver adjustment may help inform how services respond to trauma. The consideration of existing screening tools and their applicability to this population could be reviewed in light of the current findings.

Finally, further research investigating the effectiveness of intra-individual interventions, including psychoeducation, parental skills behavioural management programs, and acceptance and commitment therapy is required to establish the effectiveness of these individual treatments (Brown et al, 2013b; Brown et al, 2014). There is limited research regarding the efficacy of systemic interventions, such as systemic family therapy and peer support groups for this group. Identification of the specific contexts and presenting problems where these intra-individual and systemic interventions are most helpful would assist clinicians and service managers in offering more targeted effective treatments.

4.5 Conclusion

Caregivers and families experience significant challenges when adjusting to their child's TBI. Issues associated with these fall into three main constructs: psychosocial impact on caregiver and family functioning; adjustment, coping and resilience; and relationships and support. Further qualitative research is required to validate these constructs, and integrate them into models of parental adjustment and the development and evaluation of interventions for this population.

References

Andersson K., Bellon M., and Walker R. (2016) Parents' experiences of their child's return to school following acquired brain injury (ABI): A systematic review of qualitative studies. *Brain injury*, 8, 1-10.

Brown, F.L., Whittingham, K., Sofronoff, K., and Boyd, R.N. (2013a). Parenting a child with a traumatic brain injury: Experiences of parents and health professionals. *Brain Injury*, 27(13-14), 1570-1582.

Brown, F.L., Whittingham, K., Boyd, R.N., and Sofronoff, K. (2013b). A systematic review of parenting interventions for traumatic brain injury: child and parent outcomes. *Journal of Head Trauma Rehabilitation*, 28(5), 349-360.

Brown, F.L., Whittingham, K., Boyd, R.N., McKinlay, L., and Sofronoff, L. (2014). Improving child and parenting outcomes following paediatric acquired brain injury: a randomised controlled trial of Stepping Stones Triple P plus Acceptance and Commitment Therapy. *Journal of Child Psychology and Psychiatry*, 55(10), 1172-1183

Bruce, B., and Chisholm, J. (2001). Unintentional traumatic brain injury in children: The lived experience. *Axon*, 23, 12-17.

Carroll, C., Booth, A., and Lloyd-Jones, M. (2012). Should we exclude inadequately reported studies from qualitative systematic reviews? An evaluation of sensitivity analysis in two case study reviews. *Qualitative Health Research*, 22(10), 1425-1434.

Clark, A., Stedmon, J., and Margison, S. (2008). An exploration of the experience of mothers whose children sustain traumatic brain injury (TBI) and their families. *Clinical Child Psychology and Psychiatry*, 13, 565 – 583.

Edwards A, Elwyn G, Hood K, and Rollnick S. (2000) Judging the 'weight of evidence' in systematic reviews: introducing rigour into the qualitative overview stage by assessing Signal and Noise. *Journal of Evaluation in Clinical Practice*, 6(2), 177-84.

Fay, T.B., Yeates, K.O., Wade, S.L., Drotar, D., Stancin, T., and Taylor, H.G. (2009). Predicting longitudinal patterns of functional deficits in children with traumatic brain injury. *Neuropsychology*, 23,271-282.

Gagnon, I., Swaine, B., Champagne, F., and Lefebvre, H. (2008). Perspectives of adolescents and their parents regarding service needs following a mild traumatic brain injury. *Brain Injury*, 22(2), 161-173.

Gauvin-Lepage, J., and Lefebvre, H. (2010). Social inclusion of persons with moderate head injuries: the points of view of adolescents with brain injuries, their parents and professionals. *Brain Injury*, 24(9), 1087-1097.

Gauvin-Lepage, J., Lefebvre, H.,and Malo, D. (2015). Resilience in families with adolescents suffering from traumatic brain injuries. *Rehabilitation Nursing*, 40(6), 368-377.

Guerriere, D. and McKeever, P. (1997) Mothering children who survive brain injuries: playing the hand you're dealt. *Journal for Specialist in Pediatric Nursing*, 2(3), 105 – 115.

Jones, M., Hocking, C., and Wright- St Clair, V. (2010). Parents caregiving for children after a traumatic brain injury: structuring for security. *New Zealand Journal of Occupational Therapy*, 57(1), 4-13.

Kazdin, A.E. (1997). Parent management training: evidence, outcomes and issues. *Journal of American Academy of Child and Adolescent Psychiatry*, 36, 1249-1356.

Keenan, H.T. and Bratton, S.L. (2006). Epidemiology and outcomes of paediatric traumatic brain injury. *Developmental Neuroscience*, 28, 256-263.

Kirk, S., Fallon, D., Fraser, C Robinson, G., and Vassallo, G. (2014). Supporting parents following childhood traumatic brain injury: a qualitative study to examine information and emotional support needs across key care transitions. *Child: Care, Health and Development*, 41(2), 303-313.

Lax-Pericall, M.T., and Taylor, E. (2014). Family function and its relationship to injury severity and psychiatric outcome in children with acquired brain injury: a systematic review. *Developmental Medicine and Child Neurology*, 56(1), 19-30.

Lazarus, R. S., and Folkman, S. (1984). *Stress, appraisal and coping*. New York; Springer. Cited in Oddy, M. and Herbert, C. (2003) Intervention with families following brain injury: Evidence-based practice, *Neuropsychological Rehabilitation*, 13(1-2), 259-273

Lezak, M.D. (1986). Psychological Implications of Traumatic Brain Damage for the Patient's Family. *Rehabilitation Psychology*, 31(4), 241-250.

Martin, D.A. (1988). Children and Adolescents with Traumatic Brain Injury: Impact on the Family. *Journal of Learning Disabilities*, 21(8), 464-470.

Miles, M.B., and Huberman, M.A. (2003). *Qualitative Data Analysis: An Expanded Sourcebook (2nd Edition)*. London: Sage.

Noblit, G.W., and Hare, R.D. (1988) *Meta-Ethnography: Synthesizing Qualitative Studies (Qualitative Research Methods Series II)*. London: Sage Publications Ltd.

Oddy, M. and Herbert, C. (2003) Intervention with families following brain injury: Evidence-based practice. *Neuropsychological Rehabilitation*, 13(1-2), 259-273.

Olshanky, S. (1962). Chronic sorrow: A response to having a mentally defective child. *Journal of Social Casework*, 43, 190-193.

Rashid, M., Goez, H.R., Mabood, N., Damanhoury, S., Yager, J.Y., Joyce, A.S., and Newton, A.S. (2014). The impact of pediatric traumatic brain injury (TBI) on family functioning: A systematic review. *Journal of Pediatric Rehabilitation Medicine: An Interdisciplinary Approach*, 7, 241-254.

Ring, N., Ritchie, K., Mandava, L., and Jepson, R. (2011). A guide to synthesising qualitative research for researchers undertaking health technology assessments and systematic reviews. *NHS Quality Improvement Scotland*.

Robson, T., Ziviani, J., and Spina, S. (2005). Personal experiences of families of children with a traumatic brain injury in the transition from hospital to home. *Brain Impairment*, 6(1), 45-55.

Rosigno, C.I., and Swanson, K. (2011). Parents' experiences following children's moderate to severe traumatic brain injury: a clash of cultures. *Qualitative Health Research*, 21(10), 1413-1426.

Rosigno, C.I., Grant, G., Savage, T.A., and Philipsen, G. (2013). Parent perceptions of early prognostic encounters following children's severe traumatic brain injury: 'Locked up in this cage of absolute horror'. *Brain Injury*, 13-14, 1536-1548.

Sanders, M. (1999). Triple P-Positive Parenting Program: towards an empirically validated multilevel parenting and family support strategy for the prevention of behaviour and emotional problems in children. *Clinical Child and Family Psychology Review*, 2, 71-90

Sanders, M.R., and Kirby, J.N. (2012) Consumer engagement and the development, evaluation and dissemination of evidence-based parenting programs. *Behavior Therapy*, 43(2), 236-250

Stancin, T., Wade, S.L., Walz, N.C., Yeates, K.O., Taylor, H.G. (2008). Traumatic brain injuries in early childhood: initial impact on the family. *Journal of Developmental and Behavioural Paediatrics*, 29, 253-261

Stroebe, M.S., and Schut, H. (1999). The dual process model of coping with bereavement: Rational and description. *Death Studies*, 23(3), 197-224

Taylor, H., Yeates, K., Wade, S., Drotar, D., Stancin, T., Burant, C. (2001). Bidirectional child and family influences on outcomes on traumatic brain injuries in children. *Journal of International Neuropsychological Society*, 7, 755-767.

Trenchard, S.O., Rust, S., and Bunton, P. (2013). A systematic review of psychosocial outcomes within 2 years of paediatric traumatic brain injury in a school-aged population. *Brian Injury*, 27(11), 1217-1237.

Verhaeghe, S., Defloor, T., and Grypdonck (2005). Stress and coping among families of patients with traumatic brain injury: a review of the literature. *Journal of Clinical Nursing*, 14, 1004-1012.

Wade, S.L., Walz, N.C., Cassedy, A., Taylor, H.G., Stancin, T., Yeates, K.O. (2010) Caregiver functioning following early childhood TBI: Do moms and dads respond differently? *Neurorehabilitation*, 27, 63–72.

Walsh D, and Downe S. (2006). Appraising the quality of qualitative research. *Midwifery*, 22,108-19.

Ward, H., Shum, D., Dick, B., McKinlay, L., and Baker-Tweeney, S. (2004). Interview study of the effects of paediatric traumatic brain injury on memory. *Brain Injury*, 18, 471-495.

Webster-Stratton, C. (1990). Predictors of treatment outcome in parent training for families with conduct problem children. *Behavioural Therapy*, 12, 319.

Wharewera-Mika, J., Cooper, E., Kool, B., Pereira, S., and Kelly, P. (2016). Caregivers' voices: The experiences of caregivers of children who sustained serious accidental and non-accidental head injury in early childhood. *Clinical Child Psychology and Psychiatry*, 21(2), 268-286.

Chapter 2: Major Research Project

Experiences of care and adjustment to change in caregivers of children with autoimmune encephalitis: An Interpretative Phenomenological Analysis.

Philip Sharples

Mental Health and Wellbeing

University of Glasgow

1st Floor, Admin Building

Gartnavel Royal Hospital

1055 Great Western Road

Glasgow, G12 0XH

E-mail: p.sharples.1@research.gla.ac.uk

Word count with quotes: 8,831

Word count without quotes: 6,377

Preparation in accordance with guidelines for submission for *The European Journal of Paediatric Neurology* (see appendix V)

Plain English Summary

Experiences of care and adjustment to change in caregivers of children with autoimmune encephalitis: An Interpretative Phenomenological Analysis.

Background: Autoimmune encephalitis (AE) is a rare disease causing inflammation in the brain, often resulting in acquired brain injury. Some children may experience changes in behaviour, mental state and thinking abilities which can impair their development and relationships with others (Armangue, Petit-Pedrol, and Dalmau, 2012). A previous study found that behaviour difficulties in children with encephalitis increased parental distress and reduced their use of positive parenting strategies (Hooper et al. 2007). There has been no research on the views of parents with children who have AE.

Aims: This study explores the experiences of parents of children with AE during hospitalisation and after discharge with regard to their own adjustment and the services they received.

Methods: Participants were five parents of children diagnosed with AE attending the Paediatric Neurosciences Department in the Royal Hospital for Children, Glasgow. All were provided with a participant information sheet and gave their written consent. Each participant took part in a semi-structured interview that involved answering open ended questions about their experiences. Interviews were audio recorded and were later carefully transcribed by the researcher. Interpretative Phenomenological Analysis (IPA) was used to identify shared themes across the transcripts.

Main findings and conclusions: The analysis identified four super-ordinate themes common across the interviews: 1) uncertainty, 2) managing our recovery, 3) changes in my child, and 4) experiences of service provision. Parents reported the challenges they faced throughout their journey with their child, which were often mediated by their coping strategies and support from their family, friends and professionals. Overall, parents felt the care received in hospital was positive, despite reporting isolated incidents of feeling unsupported. A greater need for co-ordinated post-discharge support from hospital and clear communication and information sharing between services was reported. The applicability of these findings are limited due to similarities within the sample and the small number of people included. Areas for future research are recommended.

References

Armangue, T., Petit-Pedrol, M., and Damau, J. (2012). Autoimmune encephalitis in children. *Journal of Child Neurology*, 27(11), pp.1460 – 1469.

Hooper, L., Williams, W.H., Sarah, E.W., and Chua, K.C. (2007). Caregiver distress, coping and parenting styles in cases of childhood encephalitis. *Neuropsychological Rehabilitation: An International Journal*, 17(4-5), pp. 621-637.

Abstract

Background: Autoimmune encephalitis (AE) occurs in response to an antibody-mediated central nervous system disease and can lead to significant neurodisability. Prior research on family adjustment has described a reciprocal relationship between caregiver functioning, distress and clinical outcome in parents and children with encephalitis. There has been no previous research exploring the experiences of caregivers with a child with AE.

Aims: To explore the perspectives of parents and/or caregivers with a child diagnosed with AE regarding (i) their own adjustment from hospital admission to post-discharge, and (ii) their experiences of care and service provision.

Methods: A purposive sampling approach was used. Five parents of children with AE participated in a semi-structured interview exploring their experiences of caring for their child and service provision during acute care and post-discharge. Interpretative Phenomenological Analysis (IPA) was used to analyse the transcripts.

Main findings and conclusions: Four shared super-ordinate themes with related sub-themes emerged: (a) uncertainty, (b) managing our recovery, (c) changes in my child, (d) experiences of service provision. Participants reported emotional distress, often underpinned by recurrent experiences of uncertainty, and 'loss' of the previous child, and mediated by coping strategies and social support. While an overall positive experience of inpatient services was reported, parents often perceived post-discharge services as

lacking in co-ordination, communication and formal follow-up, resulting in unmet support needs. Implications and recommendations for services, practitioners and future research are discussed.

Keywords: Autoimmune encephalitis, Paediatric, Caregiver, Qualitative Research, Adjustment

1. Introduction

1.1 Autoimmune Encephalitis

Autoimmune encephalitis (AE) is an antibody-mediated central nervous system disease, which manifests when antibodies bind to cell surface proteins resulting in inflammation in the brain (Armangue, Petit-Pedrol, and Dalmau 2012). AE is an increasingly recognised cause of severe acquired brain injury (ABI). Anti-N-methyl-D-aspartate receptor (anti-NMDAR) encephalitis is the most common subtype in children, with an estimated incidence of 0.85 per million children in the UK every year (Wright et al, 2015). Clinical features can vary depending on the antibody but may include: seizures; movement disorders; and changes in behaviour, mood and cognition (Clews, Jeyasing, and Lim, 2015). Most children spend time in hospital for treatment, and may become acutely unwell with an uncertain prognosis. While many people will respond well to immunotherapy with a partial or complete recovery, some may experience a relapse or residual symptoms, and the mortality rate is estimated as 7% (Titulaer et al 2013). Hooper et al (2007) found the neurobehavioural consequences of encephalitis endure,

with greater behavioural disturbance and executive dysfunction associated with increased parental distress. Parents were also less likely to use proactive parenting strategies when experiencing higher degrees of distress. Despite this, outcome data for AE in children is somewhat lacking and there is currently no qualitative literature exploring caregivers experiences.

1.2 Family adjustment

Various stage theories have been proposed to conceptualise the process of family adjustment following significant health change in one of their members, often based on traditional stage models of bereavement (Kubler-Ross, 1969). Martin (1988) proposed that families of children with a TBI will typically progress through five stages: shock, denial, sorrow, anger and adaptation. Commonly, stage theories specify discrete stages, which family members typically navigate before they can begin to adjust and reorganise their lives. However, the application of these models in response to the psychological loss of a child with TBI is recognised as more complex and enduring due to developmental changes in the child, return to school, and a period of re-attachment or bonding to the 'new child' (Martin, 1988). Families can also experience 'chronic sorrow' (Olshansky, 1962), and recurrent grief in response to key life events. Transitions, such as education and employment, can create additional, long-term stressors for parents of children with ABI (Backhouse and Roger, 1999). Notwithstanding, the process of adjustment for caregivers of children with AE has not been investigated.

1.3 Parental experiences of children with TBI

The impact of paediatric AE on parents may be comparable to that described within the TBI literature as both involve brain impairment that is sudden and unexpected. Sharples (2016) conducted a synthesis of the qualitative research investigating parental experiences of caring for a child with a TBI post-discharge from hospital. Typically, parents experience significant psychosocial burden in response to the 'loss' of their previous child, the cognitive, behavioural and emotional sequelae of the child's injury and uncertainty over parenting their child and the future. This is often mediated by coping strategies employed by parents and the quality of support they receive from family, friends and professionals. Kirk et al (2014) found that parents of children with TBI often report an underpinning theme of uncertainty, focusing on the child's survival in acute care and the extent of the child's recovery post-discharge. The reciprocal relationship between parental adjustment and behavioural disturbance has also been widely reported in paediatric TBI (Taylor et al, 2001) and encephalitis (Hooper et al, 2007), emphasising the importance of considering parents' experiences to inform interventions, which can improve outcomes for the injured child, parental wellbeing and family functioning.

Despite these comparisons with the TBI literature there are important differences which may affect parental adjustment in AE, which warrant further investigation. Firstly, the type of brain injury in TBI and AE may differ in terms of severity, locality and diffuseness, which is likely to result in different challenges for the child and their family. Secondly, AE is a potentially relapsing and chronic condition as opposed to an injury caused by a single traumatic event (Titulaer et al, 2013). Finally, AE research is still developing and subsequently uncertainties remain regarding the specific cause in many cases. Therefore,

the unique factors associated with the disease and under-developed research related to AE may result in different adjustment experiences for parents.

1.4 Aims

- To investigate how caregivers of a child with AE perceive their own adjustment to change in their child's functioning from hospital admission to post-discharge.
- To explore the perceptions of caregivers of children with AE in relation to the services they received.

1.5 Research questions

- What are the experiences of caregivers of children diagnosed with AE between hospital admission and community discharge?
- What are the experiences of caregivers in relation to the care and support they received for themselves and for their child between hospital admission to community discharge?

2. Methods

2.1 Ethical Approval

This study was granted ethical approval by the West of Scotland Research Ethics Committee 1, and was approved by NHS Greater Glasgow and Clyde Research and Development (ref: GN15KH462) and the University of Glasgow (see appendix VII-IX).

2.2 Design

A qualitative design, using Interpretative Phenomenological Analysis (IPA) (Smith, Flowers and Larkin, 2009), was used to obtain rich descriptions of participant's understanding of their lived experience in response to particular phenomena, in this case caring for a child with AE and the service provision they received. IPA is rooted in the epistemological and ontological groundings of phenomenology, hermeneutics and idiography (Smith 2011). Phenomenology concerns the study of personal lived experiences, which is a key assumption of IPA. Hermeneutics emphasises the interpretation of these experiences, by both the participant and the researcher. Smith (2011) describes this process as a 'double hermeneutic' whereby the researcher tries to make sense of the participants' attempts to make sense of their own experiences. Finally, idiography considers the level of depth of these analyses from individual cases.

2.3 Sample

Participants were recruited through the paediatric neurosciences team in the Royal Hospital for Children (RHC), Glasgow. A purposive sampling approach was used to identify eligible caregivers. Neurologists and Neuropsychologists were asked to identify parents of children who met the inclusion criteria for this study. It was anticipated that 4-10 participants would be recruited, consistent with recommendations for professional doctorate research using IPA (Smith, Flowers, and Larkin, 2009). Small sample sizes allow for a richer, more detailed analysis of each transcript, which is in line with IPA's idiographic underpinnings. Additionally, this projected sample size was deemed feasible given the rarity of AE in children, and hence the small population of potential participants.

Eleven families met inclusion criteria and were invited to participate by the Neurologist or Neuropsychologist. Eight parents agreed to meet the researcher. The remaining three opted out, indicating that their participation would be too distressing, that family circumstances prevented their participation or they were not contactable. For unknown reasons three of the remaining eight participants did not attend their scheduled interviews and the lead researcher was unable to establish contact with them. Therefore, five parents from five families met the researcher to discuss the study further, provide informed consent and participate in the individual semi-structured interview.

2.4 Inclusion and Exclusion Criteria

Parents or primary caregivers:

- Currently in care of a child (<18 years) with a diagnosis of AE.
- Currently attending the RHC Neurosciences Department with their child or previously within the last 5 years.
- Over the age of 18
- No known learning disability or communication difficulties which may impact on their ability to consent or engage with the semi-structured interview.

2.5 Recruitment Procedures

Participants were selected on the basis of the inclusion criteria by their Neurologist or Neuropsychologist. Parents were then provided with a participant information sheet, containing details about the study (appendix X). They were asked to consent to their personal details being shared with the lead researcher, who later contacted consenting participants to discuss the study after they had opportunity to consider the participant information.

Interviews took place in the Clinical Research Facility at RHC for three appointments. Two participants were interviewed in their homes due to difficulties attending the interview venue. Participants were given opportunity to ask questions and review the participant information sheet. Once satisfied with the information they were provided with a consent form (appendix XI). It was specified that anonymity could not be guaranteed due to the

rarity of AE in children, but that quotes would be carefully selected with any identifying information removed. It was emphasised that participants were free to withdraw at any point and support would be offered if participants became distressed during the interview.

2.6 Research Procedures

General demographic information was gathered directly from participants prior to the interview and where necessary additional information was obtained from their child's hospital records with parental consent. Semi-structured interviews were used with participants based on an interview schedule designed for the purposes of this study (appendix XII). The researcher asked participants to describe their experiences at different times in their child's treatment: pre-diagnosis, acute care, diagnosis, discharge planning and post-discharge from hospital. The semi-structured interview allowed the researcher to ask open-ended questions, while retaining flexibility to ask follow-up prompting questions to explore participants' experiences further where necessary (Smith, Flowers and Larkin, 2009). Interviews lasted approximately 90 minutes, and ranged between 49 and 120 minutes. A digital voice recorder was used during the interviews, which were later transcribed verbatim by the researcher on an encrypted laptop, with all identifiable information removed. Participant names were replaced with pseudonyms and any identifiable information removed from quotations to protect confidentiality.

2.7 Data Analysis

The transcripts from the interviews were analysed using IPA, as described in Smith, Flowers and Larkin (2009). Firstly, the researcher 'immersed himself in the participants world' (Smith, Flowers and Larkin, 2009) through careful and repeated reading of the transcript. Secondly, the researcher made explanatory notes commenting on the semantic content and use of language throughout the interview. Thirdly, emergent themes were developed by identifying any patterns in the explanatory notes. Fourthly, the researcher began to create a map of the key themes emerging from the individual transcript. This process was repeated for each transcript. Finally, the researcher identified patterns and connections between transcripts to generate super-ordinate themes, which encompassed the key themes represented across all of the interviews. An experienced supervisor in qualitative methodology checked the validity of the researcher's initial analysis, through independent analysis of a sample of transcripts. Identified themes were discussed during research supervision and complete agreement was achieved.

2.8 Researcher Reflexivity

The influence of the researcher's experience and beliefs are emphasised in IPA, particularly during the interpretation of interview transcripts (Larkin, Watts, and Clifton, 2006). Consequently, in order to maintain a reflexive stance, a reflective log was maintained throughout the interviews and analysis. The researcher had no previous experience working with children with neurological impairments or their parents and no participant was known to him. This enabled the researcher to approach the interviews

with a more flexible and less biased interpretation of participants' accounts. The researcher had experience of working with adults with neurological impairments, some of whom had their injuries or illnesses in childhood. This was acknowledged during supervision, particularly when considering participants' concerns over the long-term future of their child. The researcher had regular supervision to reflect on the emotional impact from the interviews on himself, and to improve the validity and objectivity of his analysis.

3. Results

All five participants were females, aged 28 to 51 years and resident in Scotland, with four from the West coast of Scotland. Two participants were single parents. Three were full time carers, and the remaining two also worked part-time. Ages of the children ranged from 6 to 16 years, with two under the age of 12 and three of the five were males. Time since discharge from hospital ranged from 6 months to 3 years 9 months. All of the children had a confirmed diagnosis of AE; four were antibody negative and the other was diagnosed with Hashimoto's Encephalitis.

Four super-ordinate themes with associated sub-themes were identified from the transcripts (table 2.1).

Table 2.1: Super-ordinate themes and sub-themes

Super-ordinate themes	Sub-themes
Uncertainty	Causal factors
	Prognosis/condition
	The future/quality of life
Managing our recovery	Ensuring my child is safe
	Encouraging recovery/independence
	Impact on parent's wellbeing
	Coping/resilience
Changes in my child	Challenging behaviour and physical/cognitive symptoms
	Change in my child's identity
	Developmental regression and comparison to peers
	Social integration
Experiences of service provision	Inpatient care and treatment
	Post-discharge support
	School transition

3.1 Uncertainty

The significant influence of uncertainty on parents' wellbeing resonated throughout all interviews, shifting from the cause of their child's physical health condition and unclear prognosis during acute care, to the quality of life and future for their child during rehabilitation and post-discharge.

3.1.1 Causal factors

Some parents reported anxiety, confusion and guilt when considering the reasons their child had become unwell. This uncertainty was prevalent during the rapid deterioration in their child's physical health and was exacerbated by the lack of certainty from medical professionals:

“... it's so hard to get your head around because there's nothing that's set in stone because it basically tells you in the book that 90% of the time they have no idea what's caused it or why it's happened ... you want to know what it is that's caused it, that's the biggest thing that I feel like I want to know ... so you can try and prevent it in the future...” (Emma)

Conversely, some caregivers expressed certainty over the cause of their child's AE:

“...when I was trying to find the cause of how this could’ve happened and when we looked back it was because, it was since that HPV [Human Papilloma Virus] jag [injection]. And my dad said to the medical professional and they were like that no no that’s safe (pause) by hell.” (Amanda)

Amanda’s conviction about her belief was evident throughout the interview, despite reassurance to the contrary from medical professionals. Other parents considered alternative causes of the AE, such as their child’s diet.

3.1.2 Prognosis/Condition

Uncertainty over the outcome of their child’s unstable medical condition was common to all participants, particularly in acute care. Parents frequently described the impact of uncertainties over whether their child would survive:

“... when we took him up on the Wednesday and I looked at him and I thought, I just thought I was going to lose him. And when they said he was seriously ill and he needs to stay in, I wondered how seriously ill, you know...” (Diane)

Other’s also described their fear of their child acquiring a long-term disability:

“I think at the time, they thought... she was going to be like profoundly handicapped... and I just was like ‘I don’t know if I’m going to be able to deal with that... I want (name) back, that’s all I could think about, I just want my own daughter back... ’ (Beth)

Participant's recounted several traumatic experiences associated with these uncertainties during hospitalisation. These experiences were typically contextualised by their child's variable health and the inability of medical professionals to provide sufficient hope and reassurance:

"... you can sit and you can ask all these questions, but the doctors don't know a lot about it yet either, because it's still an illness that's not very common... it's like basically asking them to look into a crystal ball and tell you if everything's going to be all right, when they don't know and you don't know" (Emma)

3.1.3 Future/quality of life

Parent's frequently referred to their uncertainty regarding their child's future, particularly of relapse and their child's ability to lead a 'normal', fulfilling life. Fear for their child's future was often expressed during neuro-rehabilitation, and post-discharge and continued to impact on parents at the time of the interview:

"I'm worried sick about (name). How she's going to cope in the real world. I don't know how she's going to manage... college or get a job or anything like that. Erm, I worry, I think how's she going to cope if I'm not here?... I mean that's a big worry because she relies on me for everything." (Beth)

Participants also described concerns that their child's symptoms would re-emerge:

“... but to this day, I'll worry sick that he's going to have another bad run (seizures) and it's that, I do wait for that coming and I don't like that part. I try and be positive and think no, he's not had one for 10 months...” (Diane)

The need for certainty was also emphasised by Carol despite a context of receiving unwanted news about recovery:

“... (Neurologist) was like, 'to be honest this is as good as it's going to get basically', and we were like that's fine 'cause now I know what I'm dealing with...if you're getting told 2 years you're like, 'right well that's fine, it still gives him time to get better'. Whereas if he's not going to then I would just rather know and have that certainty...” (Carol)

3.2 Managing our recovery

All participants discussed their experiences of managing their own and their child's recovery both in hospital and post-discharge.

3.2.1 Ensuring my child is safe

The need to keep their child emotionally and physically safe during hospital and post-discharge was shared across participants. This was often expressed as a need to be by their child's side in hospital to maintain vigilance over their physical health:

"It was about a half an hour drive and it was rush hour traffic and I was looking at all the stupid drivers that don't get out the way for an ambulance... I was shouting f***ing move out the way (laughing). So that whole time I kept on looking at the machines, making sure they were alright and was just desperate to get (name) to (hospital)." (Amanda)

Others talked about the need to be strong in front of their child to protect them from the emotional turmoil parents were experiencing, particularly during hospitalisation:

"I can hold it together in front of (name), but as soon as I walk away, that's my time to just sort of breakdown, but he doesn't need to see that, he's going through enough as it is."
(Carol)

This strategy enabled them to present confidently in front of their child, and advocate on their behalf to professionals. Many parents described the ongoing need to keep their child safe upon their return home from hospital:

“We’ve had drop seizures in the house were he’s just (bang), so the stairs are really terrifying. I used to walk down the stairs in front of him all the time or behind him when he was going up the stairs.” (Diane)

The environmental adaptations and vigilance to keep her son safe within the home environment was stressed throughout Diane’s account. Her use of the metaphor “watch him like a hawk” provides a sense of ongoing apprehension and fear over the next seizure, which was referred to as “seizure watch” by another parent.

3.2.2 Encouraging recovery/independence

Most parents described the need to encourage their child’s independence, while continuing to maintain their safety. Many parents’ reflected on this delicate balance:

“Erm, and he went out for a walk last night with his girlfriend and I did not follow them. I wanted to but I didn’t... it’s still a huge, big thing for him to do as well, like be outside. So he’s done a big huge thing, I’m doing a huge, big thing by overcoming it.” (Diane)

Other caregivers highlighted their urgency in their child’s recovery and reflected on their need to adjust their own expectations:

“I’m a pusher, so... I wanted him to get better, I wanted him to behave, I wanted him to stop acting the way he was acting, and I didn’t care that he was ill. You know, it was, at that

point in time I was like 'just get better, hurry up and get better' you know and... you don't think, you're not thinking straight at the time... You just want everything to happen so fast, you just want everything back to normal..." (Emma)

Later in the interview Emma reflected that she has now become more understanding of her child's illness and less focussed on recovery, which she interpreted as positive, particularly for their relationship.

3.2.3 Impact on parent's wellbeing

Parents frequently referred to the general impact on their own wellbeing and relationships with others. While this was mainly mediated by changes in their child and uncertainty, it was also influenced by additional stressors, including income, employment, and other dependents. Many parents described feelings of shock, confusion and fear during initial hospitalisation:

"I walked into the room and there was a big load of people around (name) and she wasn't wakened and I just stood there, I think I was in shock because my sister and my mum, they were all crying but I was like, like a zombie state and I kind of looked and went "what's going on here"?" (Beth)

These experiences of shock and detachment were common and often compared to a different mode of functioning. Beth referred to herself as in "survival mode", conversely Amanda conceptualised herself as in "business mode" where she focussed on concrete

tasks. Parents also discussed the long term consequences of their child's physical condition on their own wellbeing:

"I was actually really stressed, erm, I lost weight at the time as well because of worry and I just couldn't sleep because I was waiting to hear that noise all the time (hospital machine) anyway and then when I did doze off, I would hear it and it wasn't him, it was like my own mind and things like that. Eh, that was horrific, I went to my own Doctors about that, 'cause I couldn't, I couldn't cope, I thought, I'm not going to be able to cope. How am I supposed to look after my own son, when I'm not looking after myself and I'm not sleeping and things" (Diane)

Participants reflected on their relationships with family and friends, some parents described losing contact with others, while most acknowledged becoming closer to their partner, and/or family members:

"Well surprisingly me and (husband), we became closer rather than fighting with each other, which was quite surprising 'cause it could have easily went the other way and we could have fell out over everything but we sort of worked together." (Carol)

3.2.4 Coping and resilience

All parents reported using various coping strategies to manage the emotional impact of their experiences caring for their child. Access to social support was particularly

prevalent, some needing practical support and others emotional support and reassurance from family/friends or others who understood their experiences:

“I just needed to talk about it and I went to a wee [small] carer’s course thing. And that was actually fantastic. It, you met people that you didn’t know, that had been through a lot of health issues and things like that, and they discussed it and you know, some people are worse off than yourself as well sometimes and you can talk about it, people were crying, and got to air it out, what you were going through.” (Diane)

The importance of being able to express emotions and listen to the experiences of others appeared to resonate for other parents. Many identified the need to seek out further information, while taking care that this was from reliable, trustworthy sources:

“Even online, something to read, that is a society or somebody that’s already made up, they’re only going to tell you the truth or the story so far. If you go reaching at other places, like you maybe get some quack saying... they don’t need this... medicine you can cure them with herbs and spices, do you know what I mean. So I’ve stayed away from all that and I’ve just stayed with the guidelines...” (Amanda)

Some parents reflected on the benefit of maintaining their identity as a parent in acute care:

“I was doing his wee care and stuff, washing his face and brushing his teeth. Just wee stuff like that does help because you then still feel as your part of taking care of him and

being a part of his life. Although you're not doing the main parts that you normally do and that's, that's hard like letting go. That bit, realising that somebody else has to step in to like, properly take care of him like the doctors and the nurses 'cause obviously they're better trained than I will ever be..." (Carol)

Other parents also described the importance of maintaining their relationship with their child by talking to them and using music and humour during acute care.

3.3 Changes in my child

A prominent theme throughout the interviews was changes in their child's presentation after the onset of AE.

3.3.1 Challenging behaviour and cognitive/physical symptoms

The emergence of behavioural changes was a strong theme for all parents during hospital and post-discharge. This commonly presented as aggression during acute care, often attributed to difficulties in regulating emotions, disorientation or memory impairments:

"... the only way I can describe him was like, he was like Tarzan, he had no idea what was going on round about him or who anybody was... he wasn't able to understand a conversation, you couldn't hold a conversation with him..." (Emma)

Difficulties managing changes in their child's behaviour were also common for many parents upon their return home from hospital:

"She goes off on one, you could be in the shop and say the wrong thing and she just goes "ah shut it" (angry voice)... and runs away up the street... Erm, she goes into these wee moods, it's like, you'll say to her "is everything okay?". "Just leave me alone" (angry voice), and I'm like that alright, fine then ... it's horrible. 'Cause you don't know, it's like living with two different people..." (Beth)

Some changes in their children's presentation were commonly attributed to the side-effects of their steroid or anti-epileptic medication. Diane described the impact of managing her son's seizures:

"I was scared he was going to have one (seizure) all the time and crack his head open. Erm, so we had to get a new, a door turned round the other way because I had to smash my door in. He took a bad seizure in the toilet and I was in myself with him and erm, he was behind the door and I couldn't get in to help him so had to try break (it) down..." (Diane)

Cognitive impairments also concerned many parents, particularly memory impairments and disorientation.

3.3.2 Change in my child's identity

Many parents reported experiencing a loss of their 'previous' child, often conceptualised in relation to changes in their child's cognition, behaviour, personality and emotions:

"I wasn't (coping), (crying) (pause) Sorry. I just felt like he wasn't mine. It was like, they had taken away like a child that was basically text book perfect and replaced him with a monster. It was horrible (crying)." (Emma)

Parents also reflected on the need to adjust to these changes in their child's identity upon their return home from hospital:

"... you've got to learn to adapt to the challenging behaviour that they, that they show and 9 times out of 10 when your child comes out of hospital it's not the same person. It is a different person that comes out and you've got to try and like, put, I know you still remember what they were like before, but you've got to try and love the person that's come out of hospital." (Beth)

3.3.3 Developmental regression and comparison to peers

Perceiving their child as functioning at a younger developmental age was common both in acute care and post-discharge. Some parents made comparisons between their child and their 'previous' child:

“She’s learning to swim again because she’s lost it, she thought that when you go into the water, the water goes into your ears you’ll drown... so that’s what I’m saying it was some of the things she’s like a 5 year old...” (Amanda)

Many also compared their child with other children:

“It’s like, you want to ask, you want to know, if he’s doing really well in terms of a normal child, or is he doing really well because you’re making allowances for that he was ill, you know. So it’s, it’s hard to understand if he’s averaging yet or not, just below average...” (Emma)

3.3.4 Social integration

Many parents reflected on their concerns about their child’s ability to maintain and develop friendships with their peers after discharge from hospital. Some expressed anxiety over their child’s vulnerability and experiences of bullying:

“I know that teenagers don’t have much empathy, erm but when your own child is getting, is being a victim of their, their kinda, like sick games so it is, that’s what it’s like.” (Beth)

Parents expressed a strong sense of powerlessness when trying to facilitate age-appropriate, social opportunities for their child:

“... it’s no good me going with a group of them to the pictures and going with her, do you know what I mean? It’s not the same, she’s not ready for that anyway... find it frustrating that the pals that she did have, it was a novelty at first, hanging about with the brain injured child, a lassie [girl] who’s getting attention but now they’ve just got fed up because (name) can’t do the same things as they do... so the, the ones that she did have as friends are not really there anymore... they’ve grown.” (Amanda)

Concerns over younger children’s social functioning were less prevalent for parents, however, Emma reflected on her worries about her child’s social relationships in the future:

“I’m concerned that, that, that he won’t be accepted, that’s my biggest concern, is that he’ll struggle through life socially. Erm, because obviously people are, people are cruel, eh. They don’t think about, that this person could have a mental illness, or this person could have had an illness as a child, they just, they just put them down to being stupid, you know.” (Emma)

3.4 Experiences of service provision

All parents discussed their experiences of services during their child’s inpatient treatment, post-discharge from hospital and during transition back into education.

3.4.1 Inpatient care and treatment

While all participants described their overall experiences in acute care as positive:

“... if that you needed any, any help or anything like that, the staff were always there to help you. Erm, and if you needed to speak to somebody like... the clinical nurse specialist... just said to me, that if you need to speak to me about anything just, just phone me or anything like that, so that was good.” (Beth)

Many commented on isolated events when nursing staff seemed inattentive to the child's condition or parental needs in the ward:

“There really was no staff and even sometimes, not saying they're incompetent because that's not nice but the staff were under a lot of stress and you thought I can't leave her with you in case anything happens.” (Amanda)

Amanda frequently expressed her concerns about leaving her child in the care of certain nurses because her child's condition had previously destabilised when under their care.

The importance of receiving clear, accessible information and communication from medical professionals was commonly emphasised, particularly at the point of diagnosis:

“Erm, the Doctor explained it in the office and things, I think it was Dr (name) that diagnosed him with that. And he used to show us the MRIs, brain and things like that, and you know, like where it was whiter and things ‘cause of the inflammation... it was just like we’d never heard of it before in our lives, so we had to read up on it...” (Diane)

3.4.2 Post-discharge support

Parents generally expressed positive experiences of post-discharge support, however, similar to inpatient care this was not uniform. Most parents said it was important to know they could approach the health professionals for reassurance and guidance:

“... people listen to you when you’re asking for help and they usually do give me some advice. I can go up, come in and speak to them about anything and things like that. Erm, and they’ll say just pick up the phone... if you need to speak to anybody about things. And a lot of times it’s just that reassurance.” (Beth)

Despite this, some also recognised a lack of formal, follow-up support:

“I guess we sort of feel like after discharge we were just sort of, although we could phone, we were just sort of abandoned and sort of left with this wee boy with all these problems that we didn’t fully understand our self. That was on steroids and very angry with the world and we just didn’t know what to do for the best or anything like that. I think that was, we just sorta felt you’ve got so much support in hospital then you’re out of it and you’ve not really got that there anymore, which was a big problem and a big adjustment...” (Carol)

Some parents noted the lack of outreach support and assessment from community health services:

“... his fine motor skills were affected, he couldn't hold a pencil or fork and knife and stuff like that, so he got referred to, erm, them (allied health professional) and he didn't even get an appointment with them, he just got dropped. I was like 'you've never even met him or seen him...' (Emma)

Difficulties obtaining practical support through social work were also reported by some parents:

“I'm still waiting on a letter... Don't even know what the full service is called. It's been murder, absolute murder. Social work has been a big, big let down... I dare say, if we never kept on chasing them we would still be just us in the house... they've not done anything, not linked us with anything, not gave us any information...” (Amanda)

3.4.3 School transition

Transition back to school was a key focus for many parents upon their return home from hospital. Most described a positive experience with education services, associated with the flexibility and support they provided:

“Brilliant, absolutely fantastic,... it started off like an hour a day, then it went to half days, then when they realised he wasn’t coping... he was being able to, to learn in ways that were easier for him, instead of sitting and writing with a pencil, he was getting to play with playdoh and making shapes with the playdoh rather than drawing them....” (Emma)

However, not all parents felt their child’s transition was managed effectively:

“I don’t think she was ready, I think there’s too much emphasis when a kid comes out of hospital to get them straight into education. And I think they really need a good while to, to help their brain to improve before they get pushed into doing maths and, you’re in with a big crowd of people and things like that and dealing with social situations, it was horrendous for her. And it was horrible for me ‘cause I knew I was putting her into the lion’s den because that’s what it’s like in secondary school. And a lot of the times I had to pick up the pieces...” (Beth)

Beth’s frustration over her child’s transition back into school was evident throughout her interview. She reported that her daughter was rushed back into education, experienced bullying from other pupils and emphasised the school’s inadequate knowledge and communication about her child’s illness.

4. Discussion

4.1 Impact of caring for a child with AE

Uncertainty was pervasive in most parents throughout acute care to post discharge. During the acute stage parents reported fear and anxiety associated with uncertainty about their child's prognosis. This developed over time into uncertainties over their child's future. These worries are also reported by parents of children with TBI (Kirk et al 2014). Some parents also reported ongoing feelings of anxiety, frustration and anger over their continued uncertainty about the cause their child's illness; this does not correspond well with TBI, where the cause is usually apparent. Uncertainty was exacerbated by changes in their child's behaviour, emotional and adaptive functioning, which parents often compared to a younger developmental age.

Behavioural changes had a particular impact on parental wellbeing, which is consistent with previous quantitative research (Hooper et al, 2007; Taylor et al, 2001). Some parent's described yearning for their 'old child' and needing to adapt and re-establish their relationship, which again resonates with paediatric TBI and family adjustment (Martin, 1988; Sharples, 2016). Participants frequently referred to their concerns regarding their child's social integration, and in particular to their social functioning and acceptance, as also reported in the TBI literature (Gauvin LePage and Lefebvre, 2010).

Parents reported ongoing adjustment difficulties irrespective of the length of time since hospital discharge. This was often associated with developmental challenges in

adolescence, such as transition into a new school/college, or peers surpassing their child. These experiences are consistent with the concept of 'chronic sorrow', disruption of the family life cycle stages and the ability of family members to re-adjust their roles and relationships (Carter and McGoldrick, 1999). Therefore, rather than progressing linearly through the stages of grief and adjustment parents appeared to experience ongoing grief in relation to their child's development and their ongoing concerns about the future. The dual process model (Stroebe and Schut, 1999) suggests that people oscillate between loss orientated and restoration orientated grieving, which may reflect these parent's experiences more accurately than traditional stage models of grief.

The importance of social support for many of the parents emphasises the key role of the entire family system when adjusting to the psycho-social impact associated with paediatric AE. The application of the family systems – illness model and family resilience framework to children and young people with chronic illness (Rolland and Walsh, 2006) may provide new insights into family coping and adaptation with this population. A key premise of this model is that chronic illness impacts the entire family system and the response of the family to the chronic illness is crucial. The model considers illness specific psycho-social impacts and proposes interventions to enhance resilience and strength in the system. Future research investigating the applicability of this model for family adjustment to AE may provide new theoretical insights into adjustment and development of efficacious clinical interventions.

The need to keep their child safe, while encouraging recovery and independence was an ongoing dilemma for many parents. This has been referred to as 'structuring for security' in the TBI literature (Jones, Hocking and Wright-St Clair, 2010), where families manage two interdependent processes, namely 'holding things together' and 'joining my child with

others', in order to avoid isolation and protect their child. Parents often expressed difficulties managing these processes, some reported being over-protective and limiting their child's independence, while others found they were pushing their child too far and needed to readjust their expectations of recovery. Coping strategies varied, most expressing a more problem focused approach to managing their anxiety and uncertainty, e.g. gathering information, seeking reassurance, and protecting their child. However, participant's accounts were interspersed with evidence of emotional re-appraisal and acceptance over their child's difficulties at different points in their journey. These coping strategies varied, but were largely consistent with the concepts of problem and emotion focused coping as conceptualised in Coping Theory (Lazarus and Folkman, 1984).

4.2 Experiences of service provision

Parents generally expressed positive experiences of their child's acute care, however, most described isolated incidents when their support needs were not met. Similar experiences of 'not working together' are reported in research on parents of children diagnosed with cancer in ICU (Romaniuk and Kristjanson 1995). This has important implications for staff training and development of systems to promote individualised, family centred care. A key named member of staff who can develop and review an individualised care plan with parents during their hospital stay may facilitate more collaborative working with parents.

Most parents felt that their child's diagnosis was communicated appropriately, but reported ongoing uncertainty about AE. Consequently, participants expressed the need for clear, accessible information, to reduce their uncertainties about AE, their child's

recovery and how to manage the changing needs of their child, which is consistent with parents of children with TBI (Gagnon et al, 2008). Parents varied in their preference for information, most were happy to research information on the internet and social media, while some were happy with the information provided by medical professionals. Signposting parents to appropriate forums, such as the Encephalitis Society, Child Brain Injury Trust and NHS Scotland websites can ensure parents receive accurate and reliable information.

The need for emotional support and reassurance from other people with relevant experience, (such as professionals and other parents in similar circumstances) is consistent with reports from parents of children with TBIs (Rosignano and Swanson, 2011). The co-ordination of a peer support group may enable parents to share experiences at different stages in their journey, potentially facilitating the adjustment process and reducing anxiety surrounding uncertainties for the future. The experience of abandonment was common to many parents upon discharge, which specifically reflected the lack of formal support and is consistent with the TBI literature (Kirk et al, 2014). Parents may benefit from an identified, co-ordinating professional who can provide information, liaise with other professionals and services in order to provide consistent, co-ordinated post-discharge care for the child and their family.

A need for clear communication and information sharing between health and education services was evident, as is often reported for children with TBI (Kirk et al, 2014). Ensuring teachers are appropriately informed about AE and the child's specific impairments/learning needs is important. Again, a key worker could co-ordinate interdisciplinary working to ensure prompt information sharing and training to encourage a seamless transition back into education. The importance of teachers being flexible in

adapting to the child's learning and social needs was also prominent for many parents and should be carefully considered.

Benefits of adapted parental skills training for parents of children with TBI have been demonstrated (Brown et al, 2013b). Parents of children with AE may also benefit from this approach given the similarities in changes in behaviour and emotion regulation, and concerns over the management of the child's behaviour. A secondary benefit of this group intervention would be the chance for parents to experience peer support from other parents in similar situations. Brown et al (2013a) also suggested the possible utility of acceptance and commitment therapy (ACT) to promote acceptance and readjustment to counter the potentially enduring grief and loss many families experience,. The combination of ACT with the stepping stones positive parenting intervention has demonstrated improvements for the child's behavioural and emotional outcomes and parental management strategies for parents of children with an ABI (Brown et al 2013), and could be adapted for parents of children with AE. Cognitive rehabilitation and psychological intervention has demonstrated some efficacy for improvement in cognitive functions and psychosocial symptoms for children with ABIs, and should be considered for this group due to the higher prevalence of cognitive impairment (Ross, Dorris and McMillan, 2011).

4.3 Strengths and limitations

This study is the first to explore parental experiences of caring for a child with AE and of service provision, from acute care to post-discharge. The findings may be informative for practitioners, policy makers, and academics working with this population. Given the relative rarity of AE in children, comparisons have been made with TBI literature which suggests service improvements that could benefit the AE population.

The sample size of this study is within recommendations for a doctoral study, and is consistent with the ideographic underpinnings of IPA (Smith, Flowers and Larkin, 2009) and reflects the rarity of AE in children. The interviews were typically rich with detail, often lasting ninety minutes, with clear themes resonating with the current research literature. The super-ordinate themes and sub-themes reoccurred across the interviews suggesting that data saturation appears to have been achieved.

A key limitation of qualitative and IPA studies concerns the issue of how well the identified themes generalise to the wider population. There appeared to be a balance of age, gender and time since discharge for the children with AE. However, all parents were female, of similar socio-economic and cultural backgrounds, and all were Scottish, limiting generalisability. It is also not known if those who chose not to participate had different experiences. A further limitation inherent in qualitative research and extending to this study is the subjectivity of analysis and interpretation, which can be influenced by researcher bias. Maintenance of a reflective log, frequent research supervision, and co-rating of a sample of transcripts was implemented to maintain a reflexive stance as the

researcher's own interpretations and beliefs are embraced in IPA methodology (Larkin, Watts and Clifton, 2006).

4.4 Future investigations

Further studies investigating the experiences of male parents would be interesting as the literature suggests that males can utilise different coping styles and have differing experiences of adjustment (Wade et al, 2010). It would be interesting for further research to consider both qualitative and quantitative exploration of parents' experiences in order to develop a broader understanding and improve generalisability. Further investigation of the experiences of the child with AE would provide a more comprehensive understanding of their needs.

Future research should consider the effectiveness of intervention approaches in different contexts for parents and children with AE, e.g. inpatient and post-discharge. Evaluation of family centred care plans, counselling, peer support groups, and psycho-education may be particularly helpful during the child's inpatient and neurorehabilitation care due to the increased need for information and emotional support for families. Parents may benefit from adapted parental management approaches to support them with the behavioural and emotional changes in their child and ACT may support adjustment to the potentially chronic changes in their child and family circumstances. Furthermore, research investigating the efficacy of systemic interventions for this group post-discharge from hospital, particularly focussing on strengthening resilience and coping within family systems may improve family adjustment, coping and child outcomes. Finally, evaluating the care co-ordinator role on family outcome measures with regards to facilitating key

transitions, liaising with services and providing informational and emotional support to families may be helpful.

4.5 Conclusion

This study has presented the first detailed qualitative exploration of the experience of caring for a child with AE and service provision from acute care to post-discharge. The salient experiences for parents involved enduring uncertainty, loss and persistent anxiety in response to changes in their child. Further, parents discussed their own experiences of managing recovery and coping with the demands and stressors of caring for their child. Finally, parents explored the services they received in hospital, post-discharge and their child's transition into education. Clinical implications were discussed recommending the implementation of a key co-ordinating professional both in hospital and post-discharge. Additionally, parents may benefit from provision of and signposting to information resources, and development of adapted, group based support, parental skills training and treatment for caregivers.

References

Armangue, T., Petit-Pedrol, M., and Damau, J. (2012). Autoimmune encephalitis in children. *Journal of Child Neurology*, 27(11), 1460 – 1469.

Backhouse, M., and Roger, S. (1999). The transition from school to employment for young people with acquired brain injury: Parent and student perspectives. *Australian Occupational Therapy Journal*, 46, 99-109.

Brown, F.L., Whittingham, K., Sofronoff, K., and Boyd, R.N. (2013a). Parenting a child with a traumatic brain injury: Experiences of parents and health professionals. *Brain injury*, 27(13-14), 1570-1582.

Brown, F.L., Whittingham, K., Boyd, R., and Sofronoff, K. (2013b). A systematic review of parenting interventions for traumatic brain injury: child and parent outcomes. *Journal of Head Trauma Rehabilitation*, 28(5), 349-360.

Brown, F.L., Whittingham, K., Boyd, R.N., McKinlay, L., and Sofronoff, L. (2014). Improving child and parenting outcomes following paediatric acquired brain injury: a randomised controlled trial of Stepping Stones Triple P plus Acceptance and Commitment Therapy. *Journal of Child Psychology and Psychiatry*, 55(10), 1172-1183

Clews, S., Jeyasing, S., and Lim, M. (2015). Fifteen-minute consultation: autoimmune encephalitis. *Archives of Disease in Childhood – Education and Practice Journal*, 0, 1-6

Carter, B., and McGoldrick, M. (1999). *The Expanded Family Lifecycle. Individual, family and Social Perspectives-third edition*. Allyn and Bacon: Boston.

Gagnon, I., Swaine, B., Champagne, F., and Lefebvre, H. (2008). Perspectives of adolescents and their parents regarding service needs following a mild traumatic brain injury. *Brain Injury*, 22(2), 161-173.

Gauvin-Lepage, J., and Lefebvre, H. (2010). Social inclusion of persons with moderate head injuries: the points of view of adolescents with brain injuries, their parents and professionals. *Brain Injury*, 24(9), 1087-1097.

Hooper, L., Williams, W.H., Sarah, E.W., and Chua, K.C. (2007). Caregiver distress, coping and parenting styles in cases of childhood encephalitis. *Neuropsychological Rehabilitation: An International Journal*, 17(4-5), pp. 621-637.

Jones, M., Hocking, C., and Wright-St Clair, V. (2010). Parents caregiving for children after a traumatic brain injury: structuring for security. *New Zealand Journal of Occupational Therapy*, 57(1), 4-13.

Kirk, S., Fallon, D., Fraser, C Robinson, G., and Vassallo, G. (2014). Supporting parents following childhood traumatic brain injury: a qualitative study to examine information and emotional support needs across key care transitions. *Child: Care, Health and Development*, 41(2), 303-313.

Kübler-Ross, E. (1969). *On Death and Dying*. New York: Macmillan.

Larkin, M., Watts, S., and Clifton, E. (2006). Giving voice and making sense in interpretative phenomenological analysis. *Qualitative Research in Psychology*, 3(2), 102-120.

Lazarus, R. S., and Folkman, S. (1984). *Stress, Appraisal and Coping*. New York; Springer. Cited in Oddy, M. and Herbert, C. (2003) Intervention with families following brain injury: Evidence-based practice, *Neuropsychological Rehabilitation*, 13(1-2), 259-273.

Martin, D.A. (1988). Children and Adolescents with Traumatic Brain Injury: Impact on the Family. *Journal of Learning Disabilities*, 21(8), 464-470.

Olshanky, S. (1962). Chronic sorrow: A response to having a mentally defective child. *Journal of Social Casework*, 43, 190-193.

Rolland, J.S., and Walsh, F. (2006). Facilitating family resilience with childhood illness and disability. *Current Opinion in Pediatrics*, 18(5), 527-538.

Romaniuk, D.K., and Kristjanson, L.J. (1995). The parent-nurse relationship from the perspective of parents of children with cancer. *Journal of Pediatric Oncology Nursing*, 12(2), 80-89

Roscigno, C.I., and Swanson, K.M. (2011). Parents' experiences following children's moderate to severe traumatic brain injury: A clash of cultures. *Qualitative Health Research*, 21(10), pp. 1413 – 1426.

Ross, K.A., Dorris, L., and McMillan, T. (2011). A systematic review of psychological interventions to alleviate cognitive and psychosocial problems in children with acquired brain injury. *Developmental Medicine and Child Neurology*, 53, 692-701.

Sharples, P. (2016). Exploring the lived experience of caregivers following paediatric traumatic brain injury: A meta-ethnography. *Submitted as part of the Clinical Research Portfolio*. Glasgow: University of Glasgow, 5-41

Smith, J., Flowers, P. and Larkin, M. (2009). *Interpretative Phenomenological Analysis: Theory, Method and Research*. Sage Publications: London.

Smith, J., A. (2011). Evaluating the contribution of interpretative phenomenological analysis. *Health Psychology Review*, 5(1), 9-27.

Stroebe, M.S., and Schut, H. (1999). The dual process model of coping with bereavement: Rational and description. *Death Studies*, 23(3), 197-224.

Taylor, H., Yeates, K., Wade, S., Drotar, D., Stancin, T., Burant, C. (2001). Bidirectional child and family influences on outcomes on traumatic brain injuries in children. *Journal of International Neuropsychological Society*, 7, 755-767.

Titulaer, M.J., McCracken, L., Gabilondo, I., Armangue, T. et al. (2013). Treatment and prognostic factors for long-term outcome in patients with anti-N-Methyl-D-Aspartate (NMDA) receptor encephalitis: a cohort study. *The Lancet Neurology*, 12(2), 157-165.

Wade, S.L., Walz, N.C., Cassedy, A., Taylor, H.G., Stancin, T., Yeates, K.O. (2010) Caregiver functioning following early childhood TBI: Do moms and dads respond differently? *Neurorehabilitation*, 27, 63–72.

Wright, S., Hacoen, Y., Jacobson, L., Agrawal, S., Gupta, R., Philip, S., Smith, M., Lim, L., Wassmer, E., and Vincent, A. (2015). N-methyl-D-aspartate receptor antibody-mediated neurological disease: results of a UK-based surveillance study in children. *Archives of Disease in Childhood*, 0, 1-6.

Appendices

Page

Systematic Review

Appendix I: Guidelines for submission to Developmental Neurorehabilitation 89

Appendix II: Quality Rating Appraisal Framework (Adapted from Walsh
and Downe, 2006) 96

Appendix III: Table of quality appraisal scores of included studies..... 100

Appendix IV: Example of quality weighting.....103

Major Research Project

Appendix V: Guidelines for submission to The European Journal of Paediatric
Neurology 104

Appendix VI: Major Research Project Proposal 110

Appendix VII: University of Glasgow approval letter 123

Appendix VIII: NHS Greater Glasgow and Clyde Research and Development
approval letter 124

Appendix IX: West of Scotland Research Ethics Committee approval letter 126

Appendix X: Participant Information Sheet 128

Appendix XI: Participant Consent Form 131

Appendix XII: Interview Schedule 133

Appendix XIII: Extract from Carol's (Pseudonym) transcript, pg. 20-22..... 135

Appendix I: Guidelines for submission to Developmental Neurorehabilitation

Instructions for Authors

Developmental Neurorehabilitation considers all manuscripts on condition they are the property (copyright) of the submitting author(s) and that copyright will be transferred to the Publisher if the paper is accepted. Developmental Neurorehabilitation considers all manuscripts on the strict condition that they have been submitted only to Developmental Neurorehabilitation, that they have not been published already, nor are they under consideration for publication, nor in press elsewhere. Authors who fail to adhere to this condition will be charged all costs which Developmental Neurorehabilitation incurs, and their papers will not be published.

Contributions, which may be in the form of subject reviews, peer commentary, original scientific papers, case studies, programme developments, lessons from history, ethics and law perspectives or Letters to the Editor, should be submitted online at Developmental Neurorehabilitation's ScholarOne Manuscripts site: <http://mc.manuscriptcentral.com/tpdr> .

The main text of the work should be submitted as a Microsoft Word document (.doc). All figures and tables should be submitted as separate individual files.

To facilitate anonymous review, only the article title should appear on the first page. Authors should not include their names, telephone numbers, fax numbers or e-mail addresses inside the body of the manuscript or on any figures or tables. All identifying information will be asked for during the submission process and will be kept confidential by the journal office.

The names and present affiliations of each author should be given during the submission process. One author should be designated as the corresponding author to whom proofs and offprint requests will be addressed. Full correspondence address, including telephone and fax numbers and e-mail contact, should be provided.

Authors are invited to suggest two, independent, key figures in their field, who could be approached as independent reviewers of the paper.

Papers must have an abstract, not exceeding 150 words and should include a statement of purpose, methods used, results obtained and conclusions reached. The text should be divided into numbered sections. Original papers should use headings in the order: Introduction, Methods, Results, Discussion.

All terms to be abbreviated should be spelled out at first mention with the abbreviation following immediately in parentheses. Avoid obscure abbreviation, slang, jargon and other usage that decreases clarity.

All authors must adhere to the referencing conventions used by Developmental Neurorehabilitation, to the requirement for gender-, race-, and creed-inclusive language, and to the guidelines for animal and human research (see below).

Up to six keywords should be included.

General Guidelines

Please write clearly and concisely, stating your objectives clearly and defining your terms. Your arguments should be substantiated with well-reasoned supporting evidence.

In writing your paper, you are encouraged to review articles in the area you are addressing which have been previously published in the Journal, and where you feel appropriate, to reference them. This will enhance context, coherence, and continuity for our readers.

For all manuscripts, gender-, race-, and creed-inclusive language is mandatory.

Use person-first language throughout the manuscript (i.e., persons with brain injury rather than brain injured persons).

Abstracts are required for all papers submitted, they should not exceed 150 words and should precede the text of a paper. See below for further information.

Publishing Ethics

The Editors and Taylor & Francis Group are committed to the highest academic, professional, legal, and ethical standards in publishing work in this journal. To this end, we have adopted a set of guidelines, to which all submitting authors are expected to adhere, to assure integrity and ethical publishing for authors, reviewers, and editors.

Taylor & Francis is a member of the Committee of Publications Ethics (COPE). COPE aims to provide a forum for publishers and editors of scientific journals to discuss issues relating to the integrity of their work, including conflicts of interest, falsification and fabrication of data, plagiarism, unethical experimentation, inadequate subject consent, and authorship disputes. For more information on COPE please visit <http://publicationethics.org>.

File preparation and types

Manuscripts are preferred in Microsoft Word format (.doc files). Documents must be double-spaced, with margins of one inch on all sides. Tables and figures should not appear in the main text, but should be uploaded as separate files and designated with the appropriate file type upon submission. References should be given in Council of Science Editors (CSE) Citation & Sequence format (see References section for examples).

Manuscripts should be compiled in the following order: title page; abstract; main text; acknowledgments; Declaration of Interest statement; appendices (as appropriate); references; tables with captions (on separate pages); figures; figure captions (as a list).

Title Page

A title page should be provided comprising the manuscript title plus the full names and affiliations of all authors involved in the preparation of the manuscript. One author should be clearly designated as the corresponding author and full contact information, including phone number and email address, provided for this person. Keywords that are not in the title should also be included on the title page. The keywords will assist indexers in cross

indexing your article. The title page should be uploaded separately to the main manuscript and designated as “title page – not for review” on ScholarOne Manuscripts.

Abstract

Structured abstracts are required for all papers, and should be submitted as detailed below, following the title and author's name and address, preceding the main text.

For papers reporting original research, state the primary objective and any hypothesis tested; describe the research design and your reasons for adopting that methodology; state the methods and procedures employed, including where appropriate tools, hardware, software, the selection and number of study areas/subjects, and the central experimental interventions; state the main outcomes and results, including relevant data; and state the conclusions that might be drawn from these data and results, including their implications for further research or application/practice.

For review essays, state the primary objective of the review; the reasoning behind your literature selection; and the way you critically analyse the literature; state the main outcomes and results of your review; and state the conclusions that might be drawn, including their implications for further research or application/practice.

The abstract should not exceed 150 words.

Brief Reports

This submission format is appropriate for pilot and preliminary findings as well direct replications of research findings. Papers submitted as brief reports **must be less than 10 pages** inclusive of abstract and references and are limited to no more than two tables or figures. The Results and Discussion sections should be combined under a single heading “Results and Discussion”. The Results and Discussion section should be limited to the presentation and summary of results, brief discussion of the implications of the findings, acknowledgement of the limitations, and a suggested area for future research. The title of the paper must end with “:Brief Report”

Notes on Style

All authors are asked to take account of the diverse audience of Developmental Neurorehabilitation.

Clearly explain or avoid the use of terms that might be meaningful only to a local or national audience.

Some specific points of style for the text of original papers, reviews, and case studies follow:

- Developmental Neurorehabilitation prefers US to 'American', USA to 'United States', and UK to 'United Kingdom'.
- Developmental Neurorehabilitation uses conservative British, not US, spelling, i.e. colour not color; behaviour (behavioural) not behavior; [school] programme not program; [he] practises not practices; centre not center; organization not organisation; analyse not

analyze, etc.

- Single 'quotes' are used for quotations rather than double "quotes", unless the 'quote is "within" another quote'.
- Punctuation should follow the British style, e.g. 'quotes precede punctuation'.
- Punctuation of common abbreviations should follow the following conventions: e.g. i.e. cf.
- Note that such abbreviations are not followed by a comma or a (double) point/period.
- Dashes (M-dash) should be clearly indicated in manuscripts by way of either a clear dash (-) or a double hyphen (- -).
- Developmental Neurorehabilitation is sparing in its use of the upper case in headings and references, e.g. only the first word in paper titles and all subheads is in upper case; titles of papers from journals in the references and other places are not in upper case.
- Apostrophes should be used sparingly. Thus, decades should be referred to as follows: 'The 1980s [not the 1980's] saw ...'. Possessives associated with acronyms (e.g. APU), should be written as follows: 'The APU's findings that ...', but, NB, the plural is APUs.
- All acronyms for national agencies, examinations, etc., should be spelled out the first time they are introduced in text or references. Thereafter the acronym can be used if appropriate, e.g. 'The work of the Assessment of Performance Unit (APU) in the early 1980s ...'. Subsequently, 'The APU studies of achievement ...', in a reference ... (Department of Education and Science [DES] 1989a).
- Brief biographical details of significant national figures should be outlined in the text unless it is quite clear that the person concerned would be known internationally. Some suggested editorial emendations to a typical text are indicated in the following with square brackets: 'From the time of H. E. Armstrong [in the 19th century] to the curriculum development work associated with the Nuffield Foundation [in the 1960s], there has been a shift from heurism to constructivism in the design of [British] science courses'.
- The preferred local (national) usage for ethnic and other minorities should be used in all papers. For the USA, African-American, Hispanic, and Native American are used, e.g. 'The African American presidential candidate, Jesse Jackson...' For the UK, African-Caribbean (not 'West Indian'), etc.
- Material to be emphasized (italicized in the printed version) should be underlined in the typescript rather than italicized. Please use such emphasis sparingly. n (not N), % (not per cent) should be used in typescripts.
- Numbers in text should take the following forms: 300, 3000, 30 000. Spell out numbers under 10 unless used with a unit of measure, e.g. nine pupils but 9 mm (do not introduce periods with measure). For decimals, use the form 0.05 (not .05).

Acknowledgments and Declaration of Interest sections

Acknowledgments and Declaration of interest sections are different, and each has a specific purpose.

The Acknowledgments section details special thanks, personal assistance, and dedications.

Contributions from individuals who do not qualify for authorship should also be acknowledged here. Declarations of interest, however, refer to statements of financial support and/or statements of potential conflict of interest. Within this section also belongs disclosure of scientific writing assistance (use of an agency or agency/ freelance writer), grant support and numbers, and statements of employment, if applicable.

Acknowledgments section

Any acknowledgments authors wish to make should be included in a separate headed section at the end of the manuscript preceding any appendices, and before the references section. Please do not incorporate acknowledgments into notes or biographical notes.

Declaration of Interest section

All declarations of interest must be outlined under the subheading "Declaration of interest". If authors have no declarations of interest to report, this must be explicitly stated. The suggested, but not mandatory, wording in such an instance is: The authors report no declarations of interest. When submitting a paper via ScholarOne Manuscripts, the "Declaration of interest" field is compulsory (authors must either state the disclosures or report that there are none). If this section is left empty authors will not be able to progress with the submission.

Please note: for NIH/Wellcome-funded papers, the grant number(s) must be included in the Declaration of Interest statement.

Permission

Authors are responsible for obtaining permission to reproduce copyrighted material from other sources and are required to sign an agreement for the transfer of copyright to the publisher. As an author you are required to secure permission if you want to reproduce any figure, table or extract text from any other source. This applies to direct reproduction as well as "derivative reproduction" (where you have created a new figure or table which derives substantially from a copyrighted source). Authors are required to sign an agreement for the transfer of copyright to the publisher. All accepted manuscripts, artwork, and photographs become property of the publisher.

References

References should follow the Council of Science Editors (CSE) Citation & Sequence format. Only works actually cited in the text should be included in the references. Indicate in the text with Arabic numbers inside square brackets. Spelling in the reference list should follow the original. References should then be listed in numerical order at the end of the article. Further examples and information can be found in The CSE Manual for

Authors, Editors, and Publishers, Seventh Edition. Periodical abbreviations should follow the style given by Index Medicus.

Examples are provided as follows:

Journal article: [1] Steiner U, Klein J, Eiser E, Budkowski A, Fетters LJ. Complete wetting from polymer mixtures. *Science* 1992;258:1122-9.

Book chapter: [2] Kuret JA, Murad F. Adenohypophyseal hormones and related substances. In: Gilman AG, Rall TW, Nies AS, Taylor P, editors. *The pharmacological basis of therapeutics*. 8th ed. New York: Pergamon; 1990. p 1334-60.

Conference proceedings: [3] Irvin AD, Cunningham MP, Young AS, editors. Advances in the control of Theileriosis. International Conference held at the International Laboratory for Research on Animal Diseases; 1981 Feb 9-13; Nairobi. Boston: Martinus Nijhoff Publishers; 1981. 427 p.

Dissertations or Thesis: [4] Mangie ED. A comparative study of the perceptions of illness in New Kingdom Egypt and Mesopotamia of the early first millennium [dissertation]. Akron (OH): University of Akron; 1991. 160 p. Available from: University Microfilms, Ann Arbor MI; AAG9203425.

Journal article on internet: [5] De Guise E, Leblanc J, Dagher J, Lamoureux J, Jishi A, Maleki M, Marcoux J, Feyz M. 2009. Early outcome in patients with traumatic brain injury, pre-injury alcohol abuse and intoxication at time of injury. *Brain Injury* 23(11):853-865. <http://www.informaworld.com/10.1080/02699050903283221>. Accessed 2009 Oct 06

Webpage: [6] British Medical Journal [Internet]. Stanford, CA: Stanford Univ; 2004 July 10 - [cited 2004 Aug 12]; Available from: <http://bmj.bmjjournals.com>

Internet databases: [7] Prevention News Update Database [Internet]. Rockville (MD): Centers for Disease Control and Prevention (US), National Prevention Information Network. 1988 Jun - [cited 2001 Apr 12]. Available from: <http://www.cdcnpin.org/>

Tables and Figures: Tables and figures should not be embedded in the text, but should be included as separate sheets or files. A short descriptive title should appear above each table with a clear legend and any footnotes suitably identified below. All units must be included. Figures should be completely labeled, taking into account necessary size reduction.

Captions should be typed, double-spaced, on a separate sheet. All original figures should be clearly marked with the number, author's name, and top edge indicated.

Illustrations: Illustrations submitted (line drawings, halftones, photos, photomicrographs, etc.) should be clean originals or digital files. Digital files are recommended for highest quality reproduction and should follow these guidelines:

- 300 dpi or higher
- sized to fit on journal page
- EPS, TIFF, or PSD format only
- submitted as separate files, not embedded in text files

Color Reproduction:

Color art will be reproduced in color in the online publication at no additional cost to the author. Color illustrations will also be considered for print publication; however, the author will be required to bear the full cost involved in color art reproduction.

Please note that color reprints can only be ordered if print reproduction costs are paid. Print Rates: \$900 for the first page of color; \$450 per page for the next three pages of color. A custom quote will be provided for articles with more than four pages of color. Art not supplied at a minimum of 300 dpi will not be considered for print.

Page Proofs: All proofs must be corrected and returned to the publisher within 48 hours of receipt. If the manuscript is not returned within the allotted time, the editor will proofread the article and it will be printed per the editor's instruction. Only correction of typographical errors is permitted.

Complimentary Policy and Reprints: Authors for whom we receive a valid email address will be provided an opportunity to purchase reprints of individual articles, or copies of the complete print issue. These authors will also be given complimentary access to their final article on *Taylor & Francis Online*.

Appendix II: Quality Rating Appraisal Framework (Adapted from Walsh and Downe, 2006)

Study:

Essential criteria	Specific prompt	Score
Scope and purpose		
1. Clear statement of, and rationale for, research question/aims/purposes Total score (%) =	• Clarity of focus demonstrated	
	• Explicit purpose given, such as descriptive/explanatory intent, theory building, hypothesis testing	
	• Link between research and existing knowledge	
2. Study thoroughly contextualised by existing literature Total score (%) =	• Evidence of systemic approach to literature review, location of literature to contextualise findings, or both.	
Design		
3. Method/design apparent and consistent with research intent Total score (%) =	• Rationale given for use of qualitative design	
	• Discussion of epistemological/ontological grounding	
	• Rationale explored for specific qualitative method (e.g. ethnography, grounded theory, phenomenology)	
	• Discussion of why particular method chosen is most appropriate/sensitive/relevant for research question/aims	
	• Setting appropriate	
4. Data collection strategy apparent and appropriate Total score (%) =	• Were data collection methods appropriate for type of data required and for specific qualitative method?	
	• Were they likely to capture the complexity/diversity of experience and illuminate context in sufficient detail?	
	• Was triangulation of data sources used if appropriate?	
Sampling strategy		

5. Sample and sampling method appropriate Total score (%) =	• Selection criteria detailed, and description of how sampling was undertaken	
	• Justification for sampling strategy given	
	• Thickness of description likely to be achieved from sampling	
	• Any disparity between planned and actual sample explained	
Analysis		
6. Analytic approach appropriate Total score (%) =	• Approach made explicit (e.g. Thematic distillation, constant comparative method, grounded theory)	
	• Was it appropriate for the qualitative method chosen?	
	• Was data managed by software package or by hand and why?	
	• Discussion of how coding systems/ conceptual frameworks evolved	
	• How was context of data retained during analysis?	
	• Evidence that the subjective meanings of participants were portrayed	
	• Evidence of more than one researcher involved in stages if appropriate to epistemological/theoretical stance	
	• Did research participants have any involvement in analysis (e.g. member checking)?	
	• Evidence provided that data reached saturation or discussion/rationale if it did not	
	• Evidence that deviant data was sought, or discussion/rationale if it was not	
Interpretation		
7. Context described and taken account of in interpretation Total score (%) =	• Description of social/physical and interpersonal contexts of data collection	
	• Evidence that researcher spent time 'dwelling with the data', interrogating it for competing/alternative explanations of phenomena	

8. Clear audit trail given Total score (%) =	<ul style="list-style-type: none"> Sufficient discussion of research processes such that others can follow 'decision trail' 	
9. Data used to support interpretation Total score (%) =	<ul style="list-style-type: none"> Extensive use of field notes entries/verbatim interview quotes in discussion of findings Clear exposition of how interpretation led to conclusions 	
Reflexivity		
10. Researcher reflexivity demonstrated Total score (%) =	<ul style="list-style-type: none"> Discussion of relationship between researcher and participants during fieldwork Demonstration of researcher's influence on stages of research process Evidence of self-awareness/insight Documentation of effects of the research on researcher Evidence of how problems/complications met were dealt with 	
Ethical dimensions		
11. Demonstration of sensitivity to ethical concerns Total score (%) =	<ul style="list-style-type: none"> Ethical committee approval granted Clear commitment to integrity, honesty, transparency, equality and mutual respect in relationships with participants Evidence of fair dealing with all research participants Recording of dilemmas met and how resolved in relation to ethical issues Documentation of how autonomy, consent, confidentiality, anonymity were managed 	
Relevance and transferability		
12. Relevance and transferability evident Total score (%) =	<ul style="list-style-type: none"> Sufficient evidence for typicality specificity to be assessed Analysis interwoven with existing theories and other relevant explanatory literature drawn from similar settings and studies 	

	<ul style="list-style-type: none"> • Discussion of how explanatory propositions/emergent theory may fit other contexts 	
	<ul style="list-style-type: none"> • Limitations/weaknesses of study clearly outlined 	
	<ul style="list-style-type: none"> • Clearly resonates with other knowledge and experience 	
	<ul style="list-style-type: none"> • Results/conclusions obviously supported by evidence 	
	<ul style="list-style-type: none"> • Interpretation plausible and 'makes sense' 	
	<ul style="list-style-type: none"> • Provides new insights and increased understanding 	
	<ul style="list-style-type: none"> • Significance for current policy and practice outlined 	
	<ul style="list-style-type: none"> • Assessment of value/empowerment for participants 	
	<ul style="list-style-type: none"> • Outlines further directions for investigations 	
	<ul style="list-style-type: none"> • Comment on whether aims/purposes of research were achieved 	
Total of achieved criteria = /12		

Appendix III: Table of quality appraisal scores of included studies

Paper/ criteria	Brown et al (2013a)	Bruce & Chisholm (2001)	Clark, Stedmon & Margison (2008)	Gauvin-Lepage, Lefebvre & Malo (2015)	Jones, Hocking & Wright-St Clair (2010)	Kirk et al (2014)	Robson, Ziviani Spina (2010)
Clear statement for, research question/ aims/ purposes	1	1	1	1	1	1	1
Study thoroughly contextualised by existing literature	1	1	1	1	1	1	1
Method/ design apparent, and consistent with research intent	1	1	1	1	1	0	1
Data collection strategy apparent and	1	1	1	1	1	1	1

appropriate							
Sample and sampling method appropriate	1	1	1	1	1	1	1
Analytic approach appropriate	1	1	1	1	1	1	1
Context described and taken account of in interpretation	1	1	1	1	1	1	1
Clear audit trail given	1	0	1	1	1	1	1
Data used to support interpretation	1	1	1	1	1	1	1
Researcher reflexivity demonstrated	0	0	1	0	1	0	0

Demonstration of sensitivity to ethical concerns	1	0	1	0	1	1	1
Relevance and transferability evident	1	1	1	1	1	1	1
Total	11/12	9/12	12/12	10/12	12/12	10/12	11/12

Appendix IV: Example of quality weighting

Theme: Adjustment, coping and resilience

Concept: Encouraging recovery and independence

<u>Studies describing concept</u>	<u>Theme(s)/Subtheme(s)</u>	<u>Quality rating</u>
<u>Brown et al (2013a)</u>	<u>Emotions affect parenting</u>	<u>11/12</u>
<u>Clark, Stedmon and Margison (2008)</u>	<u>Effects of the whole family</u>	<u>12/12</u>
<u>Jones, Hocking, and Wright-St Clair (2010)</u>	<u>Getting through</u> <u>Needing to be part of things</u> <u>Using knowledge</u> <u>Setting up</u> <u>Occupations with others</u>	<u>12/12</u>
<u>Kirk et al (2014)</u>	<u>Abandonment</u>	<u>10/12</u>
<u>Robson, Ziviani and Spina (2005)</u>	<u>Significant relationships</u> <u>Coping strategies</u>	<u>11/12</u>
<u>Roscigno and Swanson (2011)</u>	<u>Architect of the positive for your child</u>	<u>11/12</u>

Appendix V: Guidelines for submission to The European Journal of Paediatric

Neurology

DESCRIPTION

The European Journal of Paediatric Neurology is the official journal of the European Paediatric Neurology Society. It aims at rapid publication of high quality, original, clinical and experimental work in and relating to all aspects of paediatric neurology and paediatric neurosciences, including molecular and genetic research, and studies of animal models of relevance to human disease.

The following articles will be considered for publication: original research papers, reviews, case studies and letters to the Editor.

Original articles: The main text of original articles should generally be in the format of: Abstract, Keywords, Introduction, Materials and Methods, Results and Discussion. Authors are reminded that articles should be clear and concise and non-standard abbreviations avoided. Please provide a brief abstract and up to six keywords separately.

Reviews: The Editor welcomes review articles on topics of interest to the readers of the journal. These can range from full-length, in-depth reviews (not more than 6000 words) to mini-reviews (up to 1500 words) on topics of current interest and importance. Reviews will be subjected to the usual peer review. The organization of review material is at the discretion of the author. Please provide a brief abstract and up to six keywords separately.

Case studies: Only those case studies will be considered for publication if they present new scientific information or a new pathogenic mechanism. Case studies should be concise, and preferably not exceed 1500 words with up to five references and one illustration. They should be in the format of Introduction, Case Study and Discussion. Please include a short abstract and up to six keywords.

Letters to the Editor: Letters containing critical comment on papers recently published in the *European Journal of Paediatric Neurology* will be considered for publication in the correspondence section. Letters should not exceed one printed page (1000 words including

references, one table or one figure). At the Editor's discretion, a letter may be sent to authors of the original paper for comment and both letter and reply may be published together. Letters should have a heading and no abbreviations.

PREPARATION

NEW SUBMISSIONS

Submission to this journal proceeds totally online and you will be guided stepwise through the creation and uploading of your files. The system automatically converts your files to a single PDF file, which is used in the peer-review process.

As part of the Your Paper Your Way service, you may choose to submit your manuscript as a single file to be used in the refereeing process. This can be a PDF file or a Word document, in any format or layout that can be used by referees to evaluate your manuscript. It should contain high enough quality figures for refereeing. If you prefer to do so, you may still provide all or some of the source files at the initial submission. Please note that individual figure files larger than 10 MB must be uploaded separately.

References

There are no strict requirements on reference formatting at submission. References can be in any style or format as long as the style is consistent. Where applicable, author(s) name(s), journal title/book title, chapter title/article title, year of publication, volume number/book chapter and the pagination must be present. Use of DOI is highly encouraged. The reference style used by the journal will be applied to the accepted article by Elsevier at the proof stage. Note that missing data will be highlighted at proof stage for the author to correct.

Formatting requirements

There are no strict formatting requirements but all manuscripts must contain the essential elements needed to convey your manuscript, for example Abstract, Keywords, Introduction, Materials and Methods, Results, Conclusions, Artwork and Tables with Captions. If your article includes any Videos and/or other Supplementary material, this should be included in your initial submission for peer review purposes. Divide the article into clearly defined sections.

REVISED SUBMISSIONS

Use of word processing software

Regardless of the file format of the original submission, at revision you must provide us with an editable file of the entire article. Keep the layout of the text as simple as possible. Most formatting codes will be removed and replaced on processing the article. The electronic text should be prepared in a way very similar to that of conventional manuscripts (see also the Guide to Publishing with Elsevier). See also the section on Electronic artwork. To avoid unnecessary errors you are strongly advised to use the 'spell-check' and 'grammar-check' functions of your word processor.

Article structure

Subdivision - numbered sections

Divide your article into clearly defined and numbered sections. Subsections should be numbered 1.1 (then 1.1.1, 1.1.2, ...), 1.2, etc. (the abstract is not included in section numbering). Use this numbering also for internal cross-referencing: do not just refer to 'the text'. Any subsection may be given a brief heading. Each heading should appear on its own separate line.

Introduction

State the objectives of the work and provide an adequate background, avoiding a detailed literature survey or a summary of the results.

Material and methods

Provide sufficient detail to allow the work to be reproduced. Methods already published should be indicated by a reference: only relevant modifications should be described.

Theory/calculation

A Theory section should extend, not repeat, the background to the article already dealt with in the Introduction and lay the foundation for further work. In contrast, a Calculation section represents a practical development from a theoretical basis.

Results

Results should be clear and concise.

Discussion

This should explore the significance of the results of the work, not repeat them. A combined Results and Discussion section is often appropriate. Avoid extensive citations and discussion of published literature.

Conclusions

The main conclusions of the study may be presented in a short Conclusions section, which may stand alone or form a subsection of a Discussion or Results and Discussion section.

Appendices

If there is more than one appendix, they should be identified as A, B, etc. Formulae and equations in appendices should be given separate numbering: Eq. (A.1), Eq. (A.2), etc.; in a subsequent appendix, Eq. (B.1) and so on. Similarly for tables and figures: Table A.1; Fig. A.1, etc.

Essential title page information

- **Title.** Concise and informative. Titles are often used in information-retrieval systems. Avoid abbreviations and formulae where possible.

- **Author names and affiliations.** Please clearly indicate the given name(s) and family name(s) of each author and check that all names are accurately spelled. Present the authors' affiliation addresses (where the actual work was done) below the names. Indicate all affiliations with a lowercase superscript letter immediately after the author's name and in front of the appropriate address. Provide the full postal address of each affiliation, including the country name and, if available, the

e-mail address of each author.

- **Corresponding author.** Clearly indicate who will handle correspondence at all stages of refereeing and publication, also post-publication. **Ensure that the e-mail address is given and that contact details are kept up to date by the corresponding author.**

- **Present/permanent address.** If an author has moved since the work described in the article was done, or was visiting at the time, a 'Present address' (or 'Permanent address') may be indicated as a footnote to that author's name. The address at which the author actually did the work must be retained as the main, affiliation address. Superscript Arabic numerals are used for such footnotes.

Abstract

A concise and factual abstract is required. The abstract should state briefly the purpose of the research, the principal results and major conclusions. An abstract is often presented separately from the article, so it must be able to stand alone. For this reason, References should be avoided, but if essential, then cite the author(s) and year(s). Also, non-standard or uncommon abbreviations should be avoided, but if essential they must be defined at their first mention in the abstract itself. The abstract should not exceed 250 words.

Highlights

Highlights are mandatory for this journal. They consist of a short collection of bullet points that convey the core findings of the article and should be submitted in a separate editable file in the online submission system. Please use 'Highlights' in the file name and include 3 to 5 bullet points (maximum 85 characters, including spaces, per bullet point). You can view example Highlights on our information site.

Keywords

Immediately after the abstract, provide a maximum of 6 keywords, using American spelling and avoiding general and plural terms and multiple concepts (avoid, for example, 'and', 'of'). Be sparing with abbreviations: only abbreviations firmly established in the field may be eligible. These keywords will be used for indexing purposes.

Abbreviations

Define abbreviations that are not standard in this field in a footnote to be placed on the first page of the article. Such abbreviations that are unavoidable in the abstract must be defined at their first mention there, as well as in the footnote. Ensure consistency of abbreviations throughout the article.

Acknowledgements

Collate acknowledgements in a separate section at the end of the article before the references and do not, therefore, include them on the title page, as a footnote to the title or otherwise. List here those individuals who provided help during the research (e.g., providing language help, writing assistance or proof reading the article, etc.).

Footnotes

Footnotes should be used sparingly. Number them consecutively throughout the article. Many word processors build footnotes into the text, and this feature may be used. Should this not be the case, indicate the position of footnotes in the text and present the footnotes themselves separately at the end of the article.

Appendix VI: Major Research Project Proposal

Cover page

Module 8: Research methods

Major research project proposal

Title: Experiences of care and adjustment to change in the carers of children with autoimmune encephalopathies: An Interpretative Phenomenological Analysis.

Matriculation number: 2109024s

Date: 28/09/2015

Version: 3

Word count: 3,268

Abstract

Background: Autoimmune encephalitis (AE) is a common pathological feature of antibody-mediated central nervous system diseases. This commonly results in neurobehavioural changes, seizures, movement disorders, cognitive deficits and neuropsychiatric symptoms. While these are relatively rare disorders they can have significant and far reaching effects on both the child and their caregivers. However, there is currently no research to date exploring the experiences of caregivers of children diagnosed with autoimmune encephalitis from pre-diagnosis to post-discharge from hospital.

Aims: Investigate the perspectives of parents and/or caregivers with a child diagnosed with autoimmune encephalitis with regard to (i) their own adjustment with their child from pre-diagnosis to post-discharge, and (ii) their experiences of care and service provision.

Methods: A semi-structured interview will be used with 4-10 parents or caregivers of children diagnosed with autoimmune encephalitis. Interviews will consider the experiences of parents or caregivers from pre-diagnosis, diagnosis and treatment, discharge and post discharge. Interpretive Phenomenological Analysis (IPA) will be used to analyse the transcripts from these interviews.

Applications: Exploration of parents and caregivers experiences may highlight important themes that can be used when supporting and managing young people with autoimmune encephalopathies. We also aim to identify gaps in current service provision and additional areas of support for both children and their caregivers throughout diagnosis, treatment, and post-discharge from hospital.

Introduction

Autoimmune Encephalitis

Autoimmune encephalitis (AE) is a common pathological feature of antibody-mediated central nervous system diseases. AE manifests when antibodies bind to cell surface proteins in error, which are associated with synaptic transmission, plasticity, or neuronal excitability. AE is typically identified by its subacute onset, abnormalities detected from magnetic resonance imaging (MRI), and inflammation detected in the cerebrospinal fluid (CSF) (Vincent, 2013). People can subsequently experience changes in their mental state, focal neurological deficits or seizures (Armangue, Petit-Pedrol & Dalmau, 2012). This often results in severe syndromes, which differ depending on the antibody, however they commonly respond to immunotherapy as a first line treatment (Lancaster, Martinez-Hernandez, & Dalmau, 2011). Common forms of AE include encephalitis with anti-N-methyl-D-aspartate receptor (anti-NMDAR) antibodies, Limbic Encephalitis (LE) with VGKC-complex antibodies, and Hashimoto's Encephalopathy (HE).

The most common form of AE in children is anti-NMDAR encephalitis, with 40% of patients being under 18 years of age (Dalmau et al, 2011). Anti-NMDAR encephalitis is caused by antibodies targeting the NR1 sub-unit of the receptor in error (Dalmau et al, 2008) and an underlying ovarian teratoma in some patients. During the prodromal stage patients may typically report symptoms consistent with a viral infection, mild cognitive changes and seizures. However, within 24 hours the following key diagnostic features typically emerge: dyskinesia; decline in consciousness; and autonomic instability. Anti-NMDAR encephalitis is commonly diagnosed through testing NMDAR antibodies in serum or CSF. Typical treatments include the removal of tumours if present and immunotherapy, which the majority of patients respond well to, however treatment can often be prolonged (Dalmau et al 2011). Additionally, patients may report residual cognitive deficits, e.g. amnesia, and may experience future relapses. A multidisciplinary approach is recommended if symptoms re-emerge as this can be beneficial for both parents and families (Tham & Kong, 2012; Houtrow et al, 2012).

LE with VGKC-complex antibodies is characterised by inflammatory lesions in the limbic system, specifically the medial temporal lobes, cingulate gyrus and amygdala (Graus et al, 2004). The most common form of LE originates from leucine-rich glioma-inactivated 1 (LGI1), which is a neuronal secreted protein (Lai et al, 2010). It is somewhat rare for children to be diagnosed with LE (Armangue, Petit-Pedrol & Dalmau, 2012). Defining features of LE include hyponatraemia and Faciobrachial Dystonic Seizures (FBDS). Timely identification and appropriate treatment of FBDS can prevent unpleasant side effects to cognitive functioning (Vincent, 2013). Other symptoms of LE typically include: short-term memory loss; hallucinations; dysarthria; insomnia; confusion; and personality changes

(Andrade, Tai, Dalmau & Wennberg, 2011). Approximately 70% of patients improve with corticosteroids, plasma exchange, or intravenous immunoglobulin (Lai et al, 2010). Although, cognition will usually significantly improve following treatment, patients will often experience residual anterograde memory problems and atrophy in the hippocampus.

Hashimoto's encephalopathy (HE) also known as Steroid-Responsive Encephalopathy with Auto-immune Thyroiditis is relatively rare with an estimated prevalence of 2 in 100,000 (Mamoudjy et al 2013). Patients with HE often experience cognitive impairment leading to changes in behaviour, neuropsychiatric symptoms, seizures, focal neurological deficits, altered state of consciousness, and dystonia. Typically patients with HE will present with a high level of serum anti-thyroid peroxidase (anti TPO) antibodies and complete, or near complete neurological recovery following corticosteroid treatment (Armangue, Petit-Pedrol & Dalmau, 2012).

Although specific diagnoses can be associated with particular neurobehavioural deficits, such as the observation of disinhibited challenging behaviour in males with anti-NMDAR encephalitis, co-morbid neuropsychiatric problems are common to this group of conditions. Additionally, the pattern of onset, assessment and treatment are similar across these conditions which leads to similar burdens for family members. Prodromal symptoms typically have a rapid onset and progression and due to the relative rarity of these disorders diagnosis may be initially unclear. Most children will experience a period of time in hospital for treatment, and may become acutely unwell. While most children will respond well to immunotherapy, this can often be a lengthy treatment process and they may experience unpleasant side-effects. Children may also experience future relapses and residual symptoms following treatment such as cognitive impairment, neurobehavioural sequelae and seizures (Vincent, 2013). This may result in the child presenting differently compared with their premorbid functioning, which is likely to impact on the process of adjustment for parents and caregivers. Despite these significant and enduring effects of AE there is currently no research exploring the experiences of parents or caregivers of children with AE with regards to their own adjustment to their child and the service provision they received.

Family adjustment to chronic illness

Parsons (1958) proposed a social theory of adjustment within family systems when a member of the family becomes unwell. Chronic illness in children can disrupt the homeostasis of the family and can result in the rest of the family adopting new roles, which can cause turbulence within the family system.

A stage theory of family adjustment following diagnosis of chronic illness (Golstein & Kennet, 2002) suggests that there are four key emotional stages family members can experience:

- (I) Shock in which the family member may find it difficult to process the information they were given and display reactions ranging from apathy to over-reaction.
- (II) Denial where the family may avoid attending to the meaning of the illness and the impact it has on the present and future of the individual and the family system.
- (III) Family members begin to process the implications of the illness and possibly research more information about it. This can result in family members feeling angry, anxious, guilty or depressed.
- (IV) The family begin to adjust and acknowledge the illness and its sequelae and make necessary adaptations in response to this.

These theories have been applied to family adjustment to general chronic ill health in another family member. However, it is unclear whether they are applicable specifically for parents of children diagnosed with AE and how this may impact on the parent-child relationship.

Parental experiences of children with an acquired brain injury (ABI)

While there has been no research to date on parental experiences following AE in their child, some research has been conducted exploring parental experiences following an acquired brain injury (ABI) in their child. Roscigno and Swanson (2011) investigated the experiences of parents of children diagnosed with a moderate to severe traumatic brain injury (TBI) up to five years post injury. Four key themes emerged:

- Grateful to still have my child
- Grieving for the child I knew
- Running on nerves
- Grappling to get what my family and child deserve.

Cultural and social obstructions were also identified which largely originated from other's misunderstandings. Future transitions for young people with ABI can also create additional, future stressors for parents, such as education and future employment (Backhouse & Roger, 1999). It would be interesting to investigate whether there are parallels between the experiences of parents of a child with an ABI and those with a child with AE due to the commonalities in the sudden onset of impairment resulting from a compromised brain.

Parental experiences of children with autoimmune encephalitis

While there have been no qualitative explorations of parental experience following AE in their child, Hooper, Williams, Sarah and Chua (2007) investigated the impact of childhood encephalitis on parental coping, mood, and disciplinary strategies. They found the neurobehavioural consequences of encephalitis appear to endure over time. Higher degrees of behavioural disturbance associated with executive functioning difficulties were significantly related to increased parental distress, irrespective of the type of coping strategy used by the parents. Parents were also less likely to use proactive parental strategies when experiencing higher degrees of distress. It was recommended that parents should be supported by services, with a particular focus on the management of dysexecutive symptoms.

Aims and research questions

Aims

- To investigate how parents and/or caregivers of a child with AE perceive their own adjustment to changes in their child's functioning from pre-diagnosis to post-discharge.
- To explore the perceptions of parents or caregivers of children with autoimmune encephalitis in relation to the health service they experienced.

Research questions

- What are the experiences of parents or caregivers of children diagnosed with autoimmune encephalitis between hospital admission and community discharge, specifically in relation to parent/carer adjustment to changes in emotions, cognition and behaviour in their children?
- What are the experiences of parents or caregivers in relation to the care and support they received for themselves and for their child with autoimmune encephalitis between hospital admission to community discharge?

Plan of investigation

Design

A qualitative design, utilising a semi-structured interview will be used for this study. Interpretative Phenomenological Analysis (IPA) will be used to analyse the transcripts from the semi-structured interviews.

Participants

The participants for this study will be parents or caregivers of children diagnosed with AE. Specifically, children with the common forms of AE: anti-NMDAR Encephalitis, Limbic Encephalitis, or Hashimotos Encephalopathy will be included in the study. Parents who currently attend the Fraser of Allander Neuroscience Unit with their child in the previous 5 years will be eligible to participate. Due to the relatively low number of potential participants their children may be at various stages of treatment.

Inclusion and Exclusion Criteria

Parents or caregivers:

- In care of a child with a diagnosis of AE.
- Previously or currently attending the Fraser of Allander Neuroscience Unit with their child within the last 5 years.
- Over the age of 18
- No known learning disability or communication difficulties which may impact on the participant's ability to engage with the semi-structured interview.

Recruitment Procedures

All potential participants will be selected on the basis of the inclusion and exclusion criteria outlined above. Suitable participants will be identified and contacted by their medical consultant in the Fraser of Allander Neurosciences Unit who will provide them with a participant information sheet for this study. Parents or caregivers who express an interest in participating in the study and consent to their personal details being passed on to the lead researcher will be sent an appointment to discuss the study further and ascertain consent. All potential participants will be given an information sheet regarding the study during this meeting and a consent form. Participants will then be given the option to take the consent form away and then a further meeting would be scheduled within 7 days to finalise consent.

Research Procedures

General demographic information of consenting participants will be provided by the medical consultant to the lead researcher if potential participants have consented for this information to be shared. This will include name and address of parents, age of child and diagnosis. Participants will then be offered an appointment with the researcher for an individual, semi-structured interview lasting approximately 60-90 minutes. An interview schedule has been devised by the researcher to guide the interview. This has been developed in line with the aims of the study and relevant literature. Open-ended questions will be used in order to encourage participants to freely convey their narrative, however, follow-up probe questions will be asked to facilitate this where necessary. Parents will be asked about their experiences during the following phases of their child's illness: initial symptom onset and first medical contact; diagnosis of AE; treatment; discharge planning; and post discharge (if applicable). A digital voice recorder will be used to record the interview, which will be later transcribed by the researcher, with any identifiable information removed for confidentiality purposes.

Data Analysis

IPA will be used to analyse the data as this approach aims to obtain a detailed understanding of a person's experience of reality in order to examine a phenomenon from that person's perspective (Smith, 1996), which is consistent with the aims of this study. Transcripts will be initially analysed by the lead researcher, who will generate a list of shared themes from the interview. In order to ensure reliability of the transcript analysis the researcher will discuss themes during academic supervision and aspects of the transcription will be coded by an experienced qualitative researcher.

Participant's demographic and personal data will be stored on an NHS encrypted computer and will not leave the Fraser of Allander Unit. At no point during the research will the research team have direct access to either the participants or child health records. Audio tapes from the interview will be stored in a locked filing cabinet, in a locked room in the University of Glasgow, and transcripts will be saved on a password encrypted University of Glasgow computer. Personal information will be deleted upon the completion of the study and audio tapes and transcripts will be destroyed after ten years.

Justification of sample size

It is expected that a total of 4 to 10 participants from a potential pool of 10 who have children currently attending the Fraser of Allander Neuroscience Unit will be potentially available for this project, which is in line with current literature on IPA and qualitative studies (Smith, Flowers & Larkin, 2009). As this is primarily an exploratory study it is difficult to ascertain how many participants will consent to their involvement in this research.

Settings and Equipment

The Fraser of Allander Neurosciences Unit has recently moved to the new Royal Hospital for Children. Arrangements will be made for the interviews to be conducted on this site. Equipment required for this study includes: a digital voice recorder (DVR), a transcription pedal with software, a laptop with NHS encryption. This equipment will be provided from the University of Glasgow's existing resources.

Health and Safety Issues

Researcher safety issues

The researcher will conduct interviews alone, during normal working hours in a NHS staffed facility. The researcher will inform the relevant staff that he has arrived, left and the location of his appointments. The researcher will be familiar with the relevant health and safety protocols for NHS Greater Glasgow and Clyde and the local NHS facility where the interviews will take place.

Participant safety issues

It is possible that participants will experience distress during the interviews when asked to recount difficult emotional events in their lives. Participants will be advised of this in the consent procedure and will have the opportunity to discuss any arising issues during the debriefing process after the interviews have taken place. The interview will be stopped if the participant demonstrates clear signs of distress and the researcher will offer support at this time.

Ethical Issues

Participants will be contacted by their assigned medical consultant at the Fraser Allander Neurosciences Unit, who will provide them with a letter of invitation. It will be made clear in this communication that refusal of participation will not affect the care or service their child currently receives. Participants who express an interest in the study will then be given the

opportunity to meet with the researcher where they will be provided with further information about the study and to discuss any concerns or questions they may have. It will be made clear that participants have the right to withdraw their consent to participate in the study at any time. Due to the relative rarity of AE particular attention to maintaining the confidentiality of participants will be given. Participants will be given an anonymous code for reporting purposes and any other identifiable information will be anonymised for the final written report such as people or places. Any quotes which may identify the participants in any way will also be omitted. However, it will be made clear to potential participants during the consent process that there may be a small possibility that they may be identified from the final report.

Financial Issues

Recording equipment should not incur any additional costs as it is anticipated that this will be provided by the existing resources in the University of Glasgow. Pen and paper will be required for the interviews. Participants will not be reimbursed for travel to the NHS facility where the interviews will take place.

Suggested Timetable

Work	Deadline
MRP Final Proposal	30 th March 2015
Application for ethical approval	May 2015
Final Approved MRP Proposal	17 th July 2015
Interviews and data analysis	September 2015 – March 2016
Final Submission	July 2016

Practical Applications

Exploration of parents and caregivers experiences may highlight important themes that can be used when supporting and managing young people with autoimmune encephalopathies. We also aim to identify gaps in current service provision and additional areas of support for both children and their caregivers throughout diagnosis, treatment, and post-discharge from hospital.

Dissemination of findings

The results from this study will form part of the lead researcher's thesis as part of the Doctorate in Clinical Psychology curriculum. It is anticipated that these findings will then be written up and disseminated via a peer-reviewed scientific journal and presented in research conferences.

References

Andrade, D.M., Tai, P., Dalmau, J., Wennberg, R. (2011). Tonic seizures: a diagnostic clue of anti-LGI1 encephalitis? *Neurology*, 76, pp. 1355-1357.

Armangue, T., Petit-Pedrol, M., & Dalmau, J. (2012). Autoimmune encephalitis in children. *Journal of Child Neurology*, 27(11), pp.1460 – 1469.

Backhouse, M., & Roger, S. (1999). The transition from school to employment for young people with acquired brain injury: Parent and student perspectives. *Australian Occupational Therapy Journal*, 46, pp. 99-109.

Dalmau, J., Gleichman, A.J., Hughes, E.G., Rossi, J.E., Peng, X., Lai, M., Dessain, S.K., Rosenfeld, M.R., Balice-Gordon, R., & Lynch, D.R. (2008). Anti-NMDA receptor encephalitis: case series and analysis of the effects of antibodies. *The Lancet Neurology*, 7, 1091-1098.

Dalmau J, Lancaster E, Martinez-Hernandez E, Rosenfeld MR, Balice-Gordon R. (2011) Clinical experience and laboratory investigations in patients with anti-NMDAR encephalitis. *The Lancet Neurology*, 10, pp. 63-74.

Goldstein, G., & Kenet, G. (2002). The impact of chronic disease on the family. *Haemophilia*, 8, pp. 461-465

Graus, F., Delattre, J.Y., Antoine, J.C., Dalmau, J., Giometto, B., Grisold, W., Honnorat, J., Smitt, P.S., Vedeler, C.H., Verschuuren, J.J., Vincent, A., & Voltz, R. (2004). Recommended diagnostic criteria for paraneoplastic neurological syndromes. *Journal of Neurology, Neurosurgery and Psychiatry*, 75, pp. 1135-1140.

Hooper, L., Williams, W.H., Sarah, E.W., & Chua, K.C. (2007). Caregiver distress, coping and parenting styles in cases of childhood encephalitis. *Neuropsychological rehabilitation: An international Journal*, 17(4-5), pp. 621-637

Houtrow, A.J., Bhandal, M., Pratini, N.R., Davidson, L., Neufeld, J.A. (2012). The rehabilitation of children with anti-N-methyl-D-aspartatereceptor encephalitis: a case series. *American Journal of Physical Medicine and Rehabilitation*, 91, pp. 435-441.

Lai, M., Huijbers, M.G., Lancaster, E., Graus, F., Bataller, L., Balice-Gordon, R., Cowell, J.K., Dalmau, J. (2010). Investigation of LGI1 as the antigen in limbic encephalitis previously attributed to potassium channels: a case series. *The Lancet Neurology*, 9, pp. 776-785.

Lancaster E, Martinez-Hernandez E, Dalmau J. (2011). Encephalitis and antibodies to synaptic and neuronal cell surface proteins. *Neurology*, 77, pp. 179-189.

Mamoudjy, N., Korff, C., Maurey, H, Blanchard, G., Steshenko. D., Loiseau-Corvez, M., Husson, B., Brauner, R., Tardieu, M., & Deiva, K. (2013). Hashimoto's encephalopathy: Identification and long-term outcome in children. *European Journal of Paediatric Neurology*, 17, pp. 280-287.

Parsons, T. (1958). Definitions of health and illness in the life of American values and social structures. Cited in: Goldstein, G. & Kenet, G. (2002) The impact of chronic disease on the family. *Haemophilia*, 8, pp. 461-465

Ramritu, P.L., & Croft, G. (1999). Needs of parents of the child hospitalised with Acquired Brain Damage. *International Journal of Nursing Studies*, 36, pp. 209 – 216

Roscigno, C.I., & Swanson, K.M. (2011). Parents' experiences following children's moderate to severe traumatic brain injury: A clash of cultures. *Qualitative Health Research*, 21(10), pp. 1413 – 1426.

Smith, J.A. (1996). Beyond the divide between cognition and discourse: Using interpretative phenomenological analysis in health psychology. *Psychology & Health*, 11(2), pp. 261-271.

Smith, J., Flowers, P. & Larkin, M. (2009). *Interpretative Phenomenological Analysis: theory, method and research*. Sage Publications: London

Tham, S.L., & Kong, K.H. (2012). A case of anti-NMDAR (N-methyl-Daspartate receptor) encephalitis: A rehabilitation perspective. *Neurorehabilitation*, 30, pp. 109-112.

Vincent, A. (2013). Developments in autoimmune channelopathies. *Autoimmunity Reviews*, 12, pp. 678 – 681.

Appendix VII: University of Glasgow approval letter



Institute of Health
& Wellbeing

TMcM/CL

Philip Sharples
66 Castlandhill Road
Rosyth
KY11 2DH

4th June 2015



Dear Philip,

Major Research Project Proposal

Parents and Caregivers of Children with Autoimmune Encephalitis: Experiences from Pre-diagnosis to Post-discharge

The above project has been reviewed by your University Research supervisor and by a member of staff not involved in your project and has now been deemed fit to proceed to ethics.

Congratulations and good luck with the study.

Yours sincerely,

A handwritten signature in black ink, consisting of several horizontal strokes.

T M McMillan
Professor of Clinical Neuropsychology
Research Director



Appendix VIII: NHS Greater Glasgow and Clyde Research and Development approval letter



Administrator: Mrs Elaine O'Neill
Telephone Number: 0141 211 1743
E-Mail: elaine.o'neill2@ggc.scot.nhs.uk
Website: www.nhsggc.org.uk/r&d

R&D Management Office
West Glasgow ACH
Dalnair Street
Glasgow G3 8SW

16 December 2015

Mr Philip Sharples
Trainee Clinical Psychologist
Inst of Mental Health and Wellbeing
Gartnavel Royal Hospital
Admin Building
1055 Great Western Road
Glasgow G12 0XH

NHS GG&C Board Approval

Dear Mr P Sharples,

Study Title:	Experiences of care and adjustment to change in the carers of children with autoimmune encephalopathies: an Interpretative Phenomenological Analysis.
Principal Investigator:	Mr Philip Sharples
GG&C HB site	Royal Hospital for Children
Sponsor	NHS Greater Glasgow and Clyde
R&D reference:	GN15KH462
REC reference:	15/WS/0259
Protocol no:	V3; 28/09/15

I am pleased to confirm that Greater Glasgow & Clyde Health Board is now able to grant **Approval** for the above study.

Conditions of Approval

1. **For Clinical Trials** as defined by the Medicines for Human Use Clinical Trial Regulations, 2004
 - a. During the life span of the study GGHB requires the following information relating to this site
 - i. Notification of any potential serious breaches.
 - ii. Notification of any regulatory inspections.

It is your responsibility to ensure that all staff involved in the study at this site have the appropriate GCP training according to the GGHB GCP policy (www.nhsggc.org.uk/content/default.asp?page=s1411), evidence of such training to be filed in the site file.

2. **For all studies** the following information is required during their lifespan.
 - a. Recruitment Numbers on a monthly basis
 - b. Any change of staff named on the original SSI form
 - c. Any amendments – Substantial or Non Substantial
 - d. Notification of Trial/study end including final recruitment figures
 - e. Final Report & Copies of Publications/Abstracts

Please add this approval to your study file as this letter may be subject to audit and monitoring.

Your personal information will be held on a secure national web-based NHS database.

I wish you every success with this research study

Yours sincerely,



Mrs Elaine O'Neill
Senior Research Administrator

Cc: Prof Tom McMillan

Appendix IX: West of Scotland Research Ethics Committee approval letter

WoSRES
West of Scotland Research Ethics Service



Professor Thomas (Tom) McMillan
Professor of Clinical Neuropsychology
University of Glasgow
Institute of Mental Health and Wellbeing, University
of Glasgow
Gartnavel Royal Hospital
Administration Building, 1055 Great Western Road
G12 0XH

West of Scotland REC 1
Research Ethics
Clinical Research and Development
West Glasgow Ambulatory Care Hospital
Dalnair Street
Glasgow
G3 8SW
(Formerly Yorkhill Childrens Hospital)

Date 15 December 2015
Direct line 0141 232 1807
E-mail WoSREC1@ggc.scot.nhs.uk

Dear Professor McMillan

Study title: Experiences of care and adjustment to change in the
carers of children with autoimmune encephalopathies:
an Interpretative Phenomenological Analysis
REC reference: 15/WS/0259
IRAS project ID: 183932

Thank you for sending the documentation in full on 15 December 2015. I can confirm the REC has received the documents listed below and that these comply with the approval conditions detailed in our letter dated 03 December 2015

Documents received

The documents received were as follows:

Document	Version	Date
Participant information sheet (PIS) [Participant_information_sheet_v3_151215]	3	15 December 2015

Approved documents

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

Document	Version	Date
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Insurance Certificate_120815]		12 August 2015
Interview schedules or topic guides for participants [Interview_Schedule_V2_25092015]	2	25 September 2015

<i>Document</i>	<i>Version</i>	<i>Date</i>
Participant consent form [Participant_Consent_Form_V3_131015]	3	13 October 2015
Participant information sheet (PIS) [Participant_information_sheet_v3_151215]	3	15 December 2015
REC Application Form [REC_Form_23102015]		23 October 2015
Research protocol or project proposal [Protocol_280915_V3]	3	28 September 2015
Summary CV for Chief Investigator (CI) [Tom McMillan CV]		
Summary CV for student [Philip Sharples CV]		
Summary, synopsis or diagram (flowchart) of protocol in non technical language [Plain_English_Summary_V2_280915]	2	28 September 2015

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

15/WS/0259	Please quote this number on all correspondence
-------------------	---

Yours sincerely

Abibat Adewumi

Abibat Adewumi
Coordinators Assistant

Copy to: *Ms Emma-Jane Gault, University of Glasgow/ NHS Greater Glasgow and Clyde*
Ms Joanne McGarry, NHS Greater Glasgow and Clyde

Appendix X: Participant Information Sheet



University
of Glasgow



Phil Sharples (Trainee Clinical psychologist)

Email: p.sharples.1@research.gla.ac.uk

Institute of Health and Wellbeing

Administration Building, 1st Floor

Gartnavel Royal Hospital

1055 Great Western Road

Glasgow. G12 0XH

Experiences of care and adjustment to change in the carers of children with autoimmune encephalopathies: an Interpretative Phenomenological Analysis.

Participant information sheet

We would like to invite you to take part in the above research study. Before you decide you need to understand why the research is being done and what it would involve for you. Please take the time to read the following information carefully. Talk to others about the study if you wish. Please ask one of the researchers if there is anything that is not clear or if you would like any additional information.

Who is conducting the research?

The research is being carried out by Phil Sharples (Trainee Clinical Psychologist) and is being supervised by Prof. Tom McMillan and Dr Liam Dorris from the University of Glasgow.

What is the purpose of this study?

The aim of this research is to explore the experiences of parents or caregivers of children with a diagnosis of autoimmune encephalitis. There will be a particular focus on your own adjustment from when you first noticed the initial symptoms to when your child was discharged back home from inpatient care. Additionally, this study will aim to explore the health service you and your child received during this time.

Why have I been invited?

You have been invited as you have been identified as a parent or caregiver of a child diagnosed with a form of autoimmune encephalitis. You were identified on the basis of your child currently or previously attending the Fraser of Allander Centre for support or treatment in the last 5 years.

What does taking part involve?

Firstly if you agree for your details to be passed on, you will be contacted by Phil Sharples to arrange a suitable day and time to meet to discuss the study further. This discussion will typically last around 30 minutes and it will involve reviewing the information on this sheet and providing you with the opportunity to ask any questions that you may have. If you are happy to take part in the study after you have considered this information, Phil Sharples will first ask you to sign a consent form which you will be given a copy of to keep. You can either sign this during the initial meeting with Phil

Sharples or, if you require more time, a further appointment can be scheduled to sign the consent form if you are willing to participate.

A further meeting at the Fraser of Allander Neurosciences centre will be arranged, which will last up to one hour and thirty minutes. During this time Phil will ask you questions about your experiences of adjustment from your child's initial symptoms to post-discharge from inpatient care, in addition to the health service you and your child received at this time. This interview will be audio recorded to ensure that the information is carefully understood and recalled by the researcher.

Do I have to take part?

No, it is up to you to decide. Phil Sharples will describe the study, go through this information sheet and answer any questions you have. You are free to withdraw at any time from the study, without giving a reason. Additionally, choosing to participate or not to participate in the study will not affect the standard of care your child receives.

What will happen to my personal information?

You should also be aware that following your consent to take part in the study, and as part of your participation, the researchers may have access to some of your personal data (such as your home address and telephone number). The researchers may also obtain information about your child's age and diagnosis for information relevant to the study. This information will be stored securely on a University of Glasgow password encrypted computer and any information relevant to you or your child will be stored in line with the Data Protection Act, which means we will not share this information with others without your permission. An exception to this is if the researchers are concerned that you may be at risk of harm to yourself or others, as they are then obliged to inform the relevant agencies. This will be discussed with you first.

Personal details will be deleted after the completion of the study. Audiotapes, signed consent forms and typed transcriptions of the interview will be stored in a locked filing cabinet and in a locked room in the University of Glasgow which only the direct research team will have access to. This information will be kept for a minimum of 10 years in line with the University of Glasgow's Code of Good Practice in Research (2011). Representatives of the study Sponsor, NHS Greater Glasgow and Clyde, may also access your and your child's information, to ensure that the study is being conducted properly.

All information used for this study will be kept confidential and any identifiable information will be removed for the final report. Some anonymised quotes from the interview may be used in the final write up of this research. Please note that due to the comparatively small number of children diagnosed with autoimmune encephalitis every year it may not be possible to guarantee anonymity, however, every effort will be made to ensure this.

What will happen with the results from the study?

The findings from this study will contribute to the main researcher's qualification in Clinical psychology and therefore will be written up as a thesis, may be published in a scientific journal and presented in conferences. As stated earlier no identifiable information will be included in the final report or publication. I will also provide you with a summary of the main research findings upon completion of the write up of this study.

Who will know I am taking part in this study?

The research team and the member of staff from the Fraser Allander Neuroscience Centre who approached you about this study will know that you are taking part.

What are the disadvantages to taking part?

It is possible that during the interview you may recall memories or events that you may find distressing or difficult to talk about. However, if this feels overwhelming then you can end the interview whenever you like or request a break if you need additional time. If you continue to feel distressed after the interview then the researcher or a member of staff from the Fraser of Allander will be available to discuss this with you. If at any point in the research process the researcher is concerned about your emotional wellbeing you will be encouraged to speak to staff or relatives, or approach your GP to ensure you receive appropriate support.

The total time you will need to spend on this study, including the initial contact discussing your potential participation, is 1.5 to 2 hours. Unfortunately, we will not be able to offer you any travel expenses but where possible we will try to coincide meetings with pre-existing appointments you have at the Fraser of Allander Centre. All appointments will take place at the new Royal Hospital for Children site.

What are the possible advantages of taking part?

There are unlikely to be any direct benefits to you or your child of taking part in this study. However, it is hoped that your participation in this research may provide us with valuable information about your experiences of how your child's illness impacted on your adjustment during the various stages of treatment. We hope this information may enable health professionals become more attuned to what caregivers may be experiencing throughout treatment and consider recommendations for any additional supports which may be beneficial. Additionally, we hope this study may highlight important gaps in current service provision for carers of children with autoimmune encephalitis, and subsequently suggest possible recommendations for how this may be improved in the future.

Who has reviewed this study?

This study has been reviewed and approved by the following bodies: West of Scotland Research Ethics Committee; NHS Greater Glasgow and Clyde Research and Development; University of Glasgow.

What should I do if I have further questions?

You will be provided with a copy of this information sheet and a signed consent form if you agree to participate in this study. If you have any additional questions about this study you can contact the following:

Irene McArthur
Neurology Nurse
Royal Hospital for Children
Glasgow
G51 4TF
Telephone Number: [0141 201 0000](tel:01412010000)

Thank you for taking the time to read this information sheet.

Appendix XI: Participant Consent Form



University of Glasgow | College of Medical,
Veterinary & Life Sciences



Phil Sharples (Trainee Clinical psychologist)
Institute of Health and Wellbeing
Administration Building, 1st Floor
Gartnavel Royal Hospital
1055 Great Western Road
Glasgow. G12 0XH
Telephone: 0141 211 0607
Email: p.sharples.1@research.gla.ac.uk

Experiences of care and adjustment to change in the carers of children with autoimmune encephalopathies: an Interpretative Phenomenological Analysis.

Participant consent form

Please
initial the
BOX

I confirm that I have read and understood the information sheet dated 15/12/2015 (version 3) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	
I understand that my participation is voluntary and that I am free to withdraw at any time until the point at which the research is written up. I understand that I do not need to give a reason for withdrawing, and my medical care and legal rights or those of my child will not be affected.	
I understand that the interview I participate in will be audio recorded and that while every care will be taken to anonymise quotes, which may be included in the research paper, it cannot be guaranteed that these will remain anonymous due to the nature of this study.	
I understand that the data collected during the study may be reviewed by the study researchers, individuals from the sponsor organisation, regulatory authorities or from the NHS GG&C research and development, where it is relevant to my taking part in this research. I give permission for these individuals to access any data collected.	
I understand that if I disclose anything that causes concern for the researcher (in terms of my safety or the safety of others) during the course of the interview then they have a duty of care to report such a disclosure to the appropriate agencies.	
I agree to take part in the above study	

--	--

(Name of participant) (Date) (Signature) --

(Name of researcher) (Date) (Signature) --

(1 copy to the participant, 1 copy for your child's medical records, 1 original for the researcher)

Appendix XII: Interview Schedule

Experiences of care and adjustment to change in the carers of children with autoimmune encephalopathies: an Interpretative Phenomenological Analysis

Major Research Project –interview schedule

Pre-interview questions:

- Age of participant
- Relationship to child
- Current age of child and age at diagnosis
- Type of encephalitis: anti-NMDAR; Hashimoto's or Limbic encephalitis
- Current living circumstances of child, e.g., hospital, home etc.
- Time since diagnosis
- Time spent in hospital: longest stay and frequency of healthcare contact
- *If applicable*: time since discharge
- Any additional health problems of the child

Parental experiences during initial symptom onset, prior to diagnosis.

1. If you could think back to the early stages when you first realised (name) was unwell, could you describe to me what happened?

Specific probes

- What were the first signs that (name) was unwell that you noticed?
- What were your thoughts about this at the time?
- How did you feel?
- What did you do in response to these signs?
- At which point did you feel you needed medical advice?

Parental experiences during the diagnostic process

2. Now if you could think back the period of time when you first sought medical advice and when a diagnosis was made, could you describe to me what happened?

Specific probes

- What was their health state like at this time?
- When the diagnosis was made, what were your thoughts and feelings about this?
- What was the service you received at this time like?

Parental experiences during hospitalisation/treatment

3. Now if you could think back to the time when (name) was receiving treatment in hospital, could you please describe to me what happened during this time?

Specific probes

- What was (name) health like during this time?
- What treatment did they receive? How did you feel about this and why?

- What was the service you received at this time like?
- How were your relationships with your child and others around you at this time?

Parental experiences during discharge planning

4. Now I would like you to think about the time when medical staff discussed discharging (name) back to your care, could you describe to me what happened?

Specific probes

- How did you feel during this time?
- What were your thoughts during this time?
- How was the service you received at this time?

Parental experiences post- discharge

5. Finally I would like you to consider the time period while (name) has been back home in your care, could you describe to me how you have experienced this up until now?

Specific prompts

- Could you describe what (name) has been like during this time?
- How have you felt during this time?
- What has your relationship been like with (name) during this time?
- How does this compare to before (name) became unwell?
- What would you consider to be the greatest challenges for you during this time?
What would you consider the greatest challenges for (name) during this time?
- How were your relationships with others around you at this time?
- What was the service you received at this time like?

General probes

- *How do you feel about that?*
- *What thoughts did you have at this time?*
- *Can you tell me more about this?*
- *Can you give me an example of this?*
- *Was there anything that helped or didn't help at this time?*

Appendix XIII: Extract from Carol's (Pseudonym) transcript, pg. 20-22

Themes	Transcript	Explanatory notes
<p>Post-discharge – early stages were dreadful</p> <p>Post-discharge – increase in appetite due to steroids</p> <p>Post-discharge – behaviour difficult to control</p> <p>Post-discharge – Child not sleeping – parental fatigue</p> <p>Post-discharge – Coping – burn off energy</p> <p>Post-discharge – seizure watch – hyper-vigilance</p> <p>Post-discharge – social work tried to get help – fell into “grey area”</p>	<p>I: And in terms of post discharge, when you took (name) home, could you tell me a little bit more about how that was in the early stages?</p> <p>P: Aw, it was dreadful, it was horrible, erm, like I said he was on steroids, erm, so he was basically trying to eat everything, and I mean everything, he would try and eat a plate, with his dinner on it, he was just so hungry all the time, erm I would just stock up on like sugar free jelly and stuff and try and feed him as much of that, but he wasn't sleeping, so we weren't sleeping, he wasn't, he was just wrecking the house, pretty much 24 hours a day, seven days a week. Erm, trying to get him out, 'cause we, that point, we stayed right next to a park, so we were trying to get him up in the park as much as possible, didn't matter if it was December, didn't matter if it was wet, you can wrap him up, he can just try and burn off some energy. But he was just not for doing anything, so it was getting to a point, that we were extremely tired as well, we were struggling to be sort of, keep moving and stuff, 'cause we weren't getting any sleep, erm (name) was occasionally still having seizures at that point as well so we were constantly on seizure watch and like say my husband was back at work, so it was just mainly me and (name) erm, especially through the nights. Erm, and social work, like I said, they did try and see if they could get any sort of help involved or anything like that, erm, but (name) sorta fell in to a grey area there, he was either too sick for some services, or not sick enough for the rest of them, so there just seems to be this void that kids like (name) just sorta fall in to. You don't really get any help.</p> <p>I: So again you mentioned a couple of things there, you mentioned about the seizure watch?</p>	<p>Initially coming home from hospital described as “terrible”. She highlights that the steroids lead to an increase in his appetite and he would try to eat “everything”, sense this was difficult to control.</p> <p>Challenging behaviour – described son as wrecking the house 24 hours a day, 7 days a week, again emphasising difficulties controlling his behaviour.</p> <p>Need to help him burn off energy. Extreme tiredness – couldn't move. Emphasises that lack of sleep was a central concern at this point.</p> <p>Mum describes being on seizure watch indicating hyper-vigilance.</p> <p>Social work tried to get help but they fell within a “grey area” he was too “sick” for some services and not “sick” enough for others.</p>

<p>Post-discharge – seizures scary need to promptly respond</p> <p>Post-discharge – Seizures still scary but reduced</p> <p>Post-discharge – processing what has happened</p> <p>Post-discharge – Uncertainty over where to start</p> <p>Post-discharge – conflict, glad to have son back but not the same child.</p> <p>Post-discharge – difficulties distinguishing between a seizure and muscle movement disorder</p>	<p>Can you tell me a little bit about how that was for you?</p> <p>P: Erm, well, even to this day, if he has a seizure, it is scary and you're like, you just sort of, you have a wee moment of 'shit, here we go again', before you need to, you spring in to action kinda thing. So, even just now, it's scary, so at that point it was a lot, lot more scary. Erm, because he was just out of hospital, we were just, think we were still trying to get our heads round the fact that he'd been in hospital for that length of time in the first place. To then be at home and dealing with everything, you're like, you just, you just don't really know where to sorta start with some stuff when you're like, you're just completely at a loss to be honest, because again, you're glad you've got you're wee boy back, but he's not that same wee boy, he's having seizures, that scare, that scare yourself like so much it's unbelievable, plus he was also hallucinations aswell. Erm, and I think at that point, he still had a bit of a muscle movement disorder so sometime trying to distinguish between a seizure and a muscle movement was quite hard aswell.</p>	<p>Emphasis on the fear associated with the seizures and continued effect on mum and subsequent need to be ready to promptly respond. Indications that while this is still scary it has improved over time.</p> <p>They were trying to come to terms and process what had happened, length of time in hospital and also deal with the sequelae of his illness. Feeling unsupported and uncertain about what to do.</p> <p>Conflict between positives of having your son back and realising he's not the same boy – hallucinations; seizures and muscle movement disorder.</p>
--	---	---