

UNIVERSITY OF LIVERPOOL

Stress and its Covariates in Carers of Children Newly Diagnosed with Epilepsy

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August 2012

Submitted in accordance with the requirements of the
University of Liverpool for the degree of Master of
Philosophy.

Abstract

Introduction: Stress is a complex concept involving a wide range of interplaying factors implicated. Chronic paediatric disease has been strongly associated with increased stress levels in parents and carers. Previous studies of carers of children with established epilepsy have also exhibited this trend when compared to healthy matched controls. The number of carers and the degree of stress experienced has a wide breadth in the existing literature, with a paucity of information relating to the period of initial presentation of seizures and diagnosis.

Methods: This is a prospective cohort study examining stress and other covariates associated with a new diagnosis of paediatric epilepsy. Recruitment occurred at first presentation to Alder Hey Children's Hospital subject to satisfying the selection criteria- primarily a child aged between 0-16 years with a suspected seizure disorder. Cohorts were retrospectively allocated following a diagnosis being made or not. The outcome measure was an 11 item questionnaire battery of which 9 were completed by the primary carer and 2 completed by the child, depending on their age. Each questionnaire was focused specifically on stress or another associated covariate. Data collection occurred at time of presentation and recruitment and a follow up approximately 6 months later, following a diagnosis having been made.

Non-parametric analyses ensued to allow for skew in the data and investigate the differences between the 2 cohorts. Statistical tests such as Mann-Whitney U tests and Spearmans rank correlations were used to analyse whether any differences observed were significant or not and to determine the strength of the correlations between stress and the other measured covariates.

Results: Of the 196 carers approached 88 were consented into the study. Although follow-up appointments are still occurring at time of writing, to date 39 complete matched data-sets have been collected with a mean time to follow-up of 6.4 months. At time of presentation no significant differences in stress and its covariates between the epilepsy and non-epilepsy cohorts based upon Mann-Whitney U analysis. However, despite this 59% of the carers regardless of their child's following diagnosis were exhibiting greater than normal levels of stress. At the second data collection point, following diagnosis, significantly higher stress levels were found in the epilepsy cohort $p=0.040$. This group of carers also demonstrated significantly higher levels of mental health concerns, difficult child behaviour and dysfunctional coping strategies.

The degree of association found between stress and the covariates was highly variable with correlation coefficients ranging from $r = -0.50$ and 0.50 for child perceived quality of life and carer mental health respectively, to $r=0.08$ and 0.13 for carer's locus of control and carer needs.

Discussion: As with the existing literature, these results suggest that a new diagnosis of paediatric epilepsy is a significant cause of stress to carers. However stress also appears to be associated with the uncertainty and experience of the pathway from first seizure event to attendance at outpatient neurology clinics. Child centred variables such as frequency of seizure events, child quality of life and child behaviour were found to be among the covariates most strongly correlated with carer stress. Future work hopes to further delineate the relationship with stress and its associated factors within the context of a new diagnosis of paediatric epilepsy. This may be achieved by larger sample sizes allowing a valid change over time analysis to be conducted.

Acknowledgements:

I'd like to take this opportunity to thank my supervisors: Dr Andrew Curran and Dr Kevin Southern for their continuous support and guidance throughout the duration of this year.

I'm additionally extremely grateful to Dr Steven Lane of the Biostatistics Department at the University of Liverpool, for all his help and patience in the role of statistical advisor for this work.

Furthermore, I'd like to extend my gratitude to all of the staff, both in the Institute of Child Health and the Neurology Department at Alder Hey Children's Hospital, for welcoming me into their respective teams and being a constant source of assistance and encouragement, in particular to Cath Denny and Norma McKnight.

Most importantly, I'm extremely thankful to all of the children and families who graciously gave up their time to take part in this work. Without them it would not have been possible.

Nomenclature:

AEDs	Anti Epileptic Drugs
CASP	Critical Appraisal Skills Programme
CBCL	Child Behaviour Checklist
DVLA	Driver and Vehicle Licensing Agency
EEG	Electroencephalogram
FAAR	Family Adjustment and Adaptability Response
FACES IV	Family Adaptability and Cohesion Scale
FBII	Family Burden of Injury Interview
FNS	Family Needs Survey
FSS	Family Support Survey
GAS	General Adaptation Syndrome
GHQ	General Health Questionnaire
GMC	General Medical Council
HRLoC	Health Related Locus of Control
HRQOL	Health Related Quality of Life
LoC	Locus of control
NICE	National Institute of Clinical Excellence
PedsQL	Paediatric Quality of Life Inventory
PIP	Paediatric Inventory for Parents
PIP-f/ PIP-d	Paediatric Inventory for Parents –frequency and –difficulty domains.
PSI	Parenting Stress Index
QRS	Questionnaire on Resources and Stress
RLoC	Rotter's Locus of Control
SDQ	Strengths and Difficulties Questionnaire
SIGN	Scottish Intercollegiate Guideline Network
SIP-A –C	Self Image Profile –for Adolescents –For Children
SSSQ	Semi-Structured Stress Questionnaire
SUDEP	Sudden Unexplained Death in Epilepsy
TBI	Traumatic Brain Injury
WHO	World Health Organisation

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Chapter 1: A Structured Review of Family Stress Theory

The topic of this work is focused upon factors influencing stress on a family environment as a result of a new diagnosis of paediatric epilepsy. A solid knowledge of the origins, definitions and development surrounding the concept of stress is therefore crucial to the understanding and undertaking of this research. As a result, a detailed literature review of the progression of stress and the associated theories as applied to a family context has been carried out and detailed below.

1.1 Stress: Background and Definitions

1.1.1 Stress

The term stress was derived from the Latin *stringere*, meaning 'to draw tight' (Rosch, 2011). Stress in its original meaning was used within the field of physics to describe elasticity, i.e. the property of a material that allows it to resume its original size and shape after having been compressed or stretched by an external force. It wasn't until the late 1930's that it began to be used within a biological context. One of its first uses was within the fields of medicine and psychology by Hans Selye, the esteemed Canadian physician, who defined it as 'the non-specific response of the body to any demand for change' (Baumann and Turpin, 2010). Following this, he also set forward one of the first models of stress named the General Adaptation syndrome (GAS) which he divided into 3 'reactionary' phases:

1. The stage of most rapid adaptation and alertness. This is in order to maintain function in the acute setting
2. Period of endurance and habituation
3. Adjustment back to baseline levels

These stages were initially designed to provide an organised framework for the physiological changes he noted during his experiments into stress. His research was primarily conducted on mouse models but subsequent work has described the characteristic stress response in humans, known colloquially as the fight or flight response. This response is regulated by two neuro-endocrine systems: 1) Hypothalamic-Pituitary-Adrenal axis and 2) Sympatho-Adrenal Medullary axis. The relationship between these two pathways and their biochemical interdependence cause a range of physiological signs (illustrated in Figure 1) as well as the psychological components of the stress response. A potential construct describing the interactions between the physiological and psychological elements of the stress is the psychophysiological stress response model (represented in Figure 2)

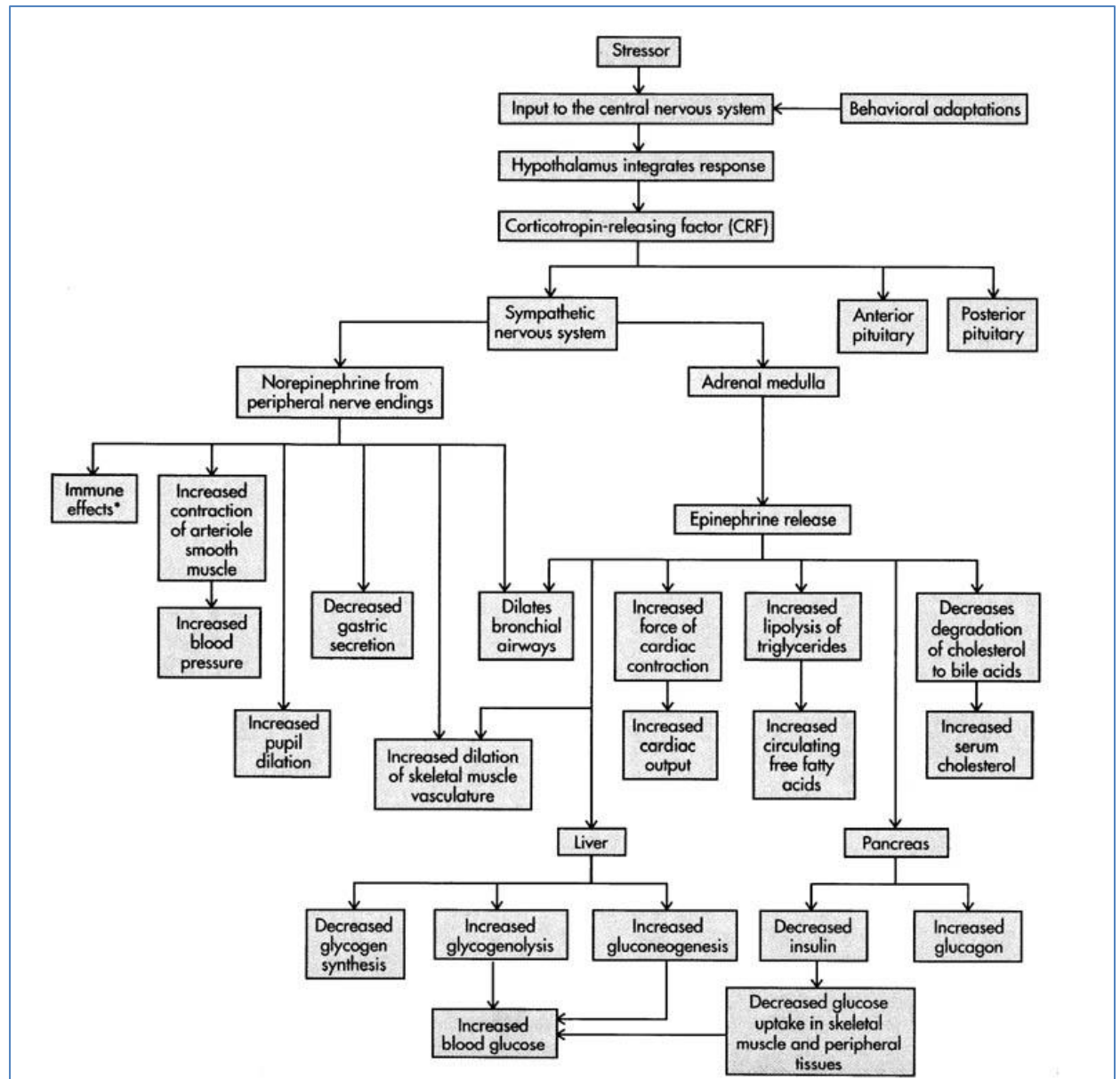


Figure 1: A Diagram illustrating the Physiological Response to Stress (Negrao et al., 2000)

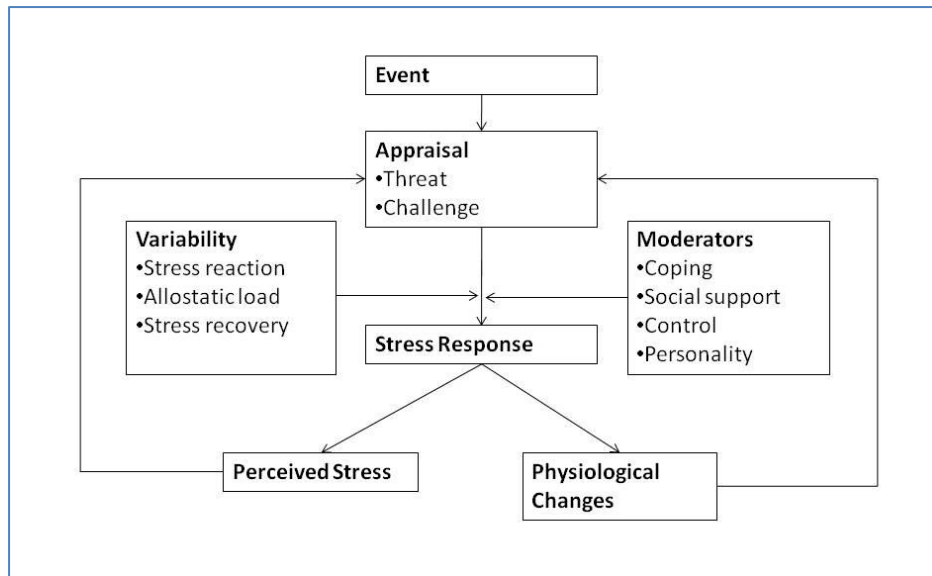


Figure 2: An adapted illustration of the psycho-physiological stress response (Ogden, 2007)

In the subsequent decades the concept of stress with regard to human biology, has greatly evolved. A more current, working description of stress is: ‘a state of tension resulting from factors that alter an existing equilibrium’ (Sanderson, 2004). This definition aims to encompass and acknowledges both the physiological changes associated with stress as well as the internal psychological response.

1.1.2 Stressors

Following the introduction of stress as a widely used concept, the term stressor was developed to refer to the stimulus which provokes a change from the existing equilibrium (Baumann, 2010). It is relevant to note that they are considered neutral and lead to stress when they necessitate change (Boss, 2002). These stressors may take a variety of forms, but are considered to be predominantly environmental in nature e.g. adverse life experiences. The stimuli are perceived by the individual to exceed their adaptive/coping capacity (See coping section below) and the input from the psychological component helps to exacerbate a stress response (Negrao et al., 2000).

Within a paediatric epilepsy setting there are likely to be a variety of different potential stressors in play, therefore as fully described in the methods, a multiple questionnaire based design and a multivariate analysis is used in order to help identify as many of these possible influential factors as possible.

1.1.3Coping

As previously stated, when using the above definitions, both the stressor and the stress itself are considered to be neutral factors. What often determines the nature of the event/change is ultimately the manner in which it is dealt with.

Therefore heavily intertwined with stress is the concept of coping. A popular definition describes it as 'the constantly changing cognitive and behavioural efforts to manage the specific external or internal demands that are appraised as taxing or exceeding the resources of the person.(Donnellan et al., 2006)' When it is constructive it can play an important factor in ones overall well-being and equally, when destructive, can have negative psychological or physical sequelae.

Associated with the process itself, are various recognised coping resources and strategies. These are respectively described as: the assets used to manage the distress caused by the stressor, and the activities/behaviours for getting/using the resources in order to restore equilibrium. Both of which, are thought to aid in reducing stress (Ogden, 2007).

Although the literature is vast and with some degree of discord, broadly speaking coping strategies can be divided into 2 key categories: problem focused and emotion focused coping (Billings and Moos, 1981). This is also known as instrumentality-emotionality in the literature.

With regard to this research, understanding a family's coping mechanisms gives an important insight into the way in which they perceive and approach new and potentially stressful circumstances. Furthermore, it has been well documented that in paediatric trauma, constructive family coping strategies can have a significant effect on child health outcomes(Dunst and Trivette, 1986).

1.2 The Family and Family Stress Theory

1.2.1 Family

To fully appreciate the concept of family stress, it must first be clarified exactly what is meant by a family. In legal terms a family is defined as: 'A group of individuals who share ties of blood, marriage, or adoption; a group residing together and consisting of parents, children, and other relatives by blood or marriage; a group of individuals residing together who have consented to an arrangement similar to ties of blood or marriage.'

This definition encompasses many possible family organisations, reflecting changes in modern times. In the last century alone, the family unit has greatly changed: from the classical nuclear family to a wide variety of different family structures, which may be loosely divided into 5 distinct types (Bengtson, 2001):

- Nuclear Families: two parents (often married) with their children. All present in the same home.
- Single Parent Families: One parent in charge of his/her children.
- Step Families: Families with a parent or parents who also has children from a prior relationship. These children may or may not be living within the same home.
- Extended families: Similar structure to a nuclear family but with the addition of other family members, most often grandparents.
- Adopted families: Families, often nuclear in nature, who have adopted non-biological children into their unit.

Regardless of their differing structures, each family is subject to a range of roles and interactions (Cox and Paley, 1997). As suggested by family systems theories, each individual contributes to the formation of a group and as such there multiple ties connecting every component person to each other, in addition to the interaction of the unified group with their environment (Cox and Paley, 1997). Considering the family as a group it is apparent that it is via these links between members that a stressor affecting a specific member may be transmitted and experienced by other members within the family unit.

1.2.2 Family Stress

Following the initial introduction of stress as a psychological concept, family stress later became an area of interest in its own right. It can be formally defined as the state which arises as a result of an actual or perceived imbalance of the demand-capability ratio within a family's functioning. It is used to help explain the effect of stress upon a family dynamic and to understand the coping mechanisms developed by families in order to counter the sources of the stress and its associated hardships.

To provide a clear structure to the complex concepts and processes relating to family stress these theories have been developed into formal models. This allows greater clarity and easier application to real-life scenarios.

Since its inception in 1930's, primarily carried out by graduate students working for the psychologist Angell (McCubbin, 1979), the early models of family stress have been altered and adapted in response to a greater research and knowledge base as illustrated by the timeline below:

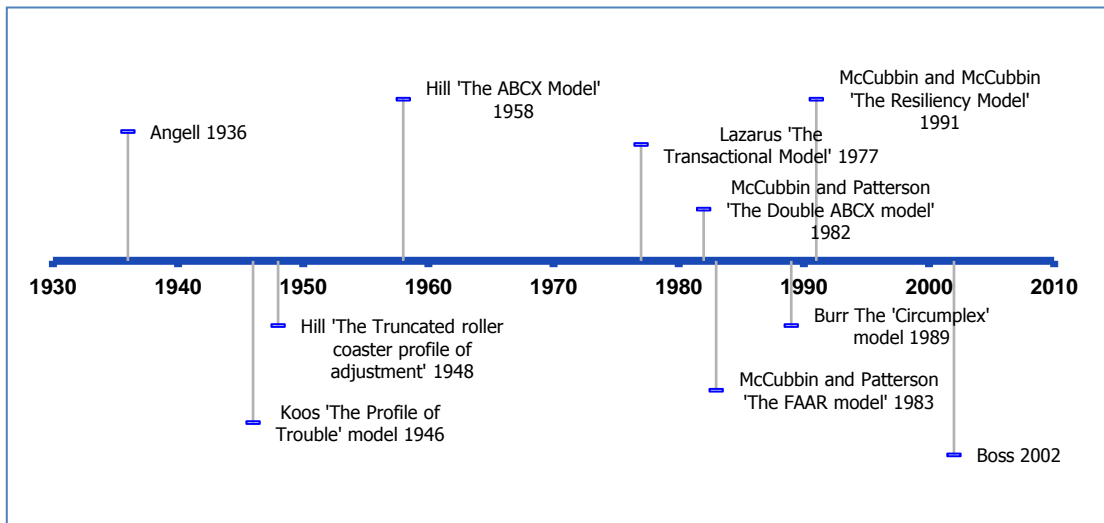


Figure 3: A Timeline of the Progression in Family Stress Theory. Adapted from (Gauthier-Weber, 2011)

1.3 Models of Family Stress

Many constructs have been developed in order to illustrate different aspects of family stress. This study aims to identify factors influencing stress in families as a result of paediatric epilepsy. Therefore awareness of the different models as well as an understanding of their relative strengths and weaknesses is essential to the study design. Described below are some of the principal models regarding family stress.

1.3.1 The ABCX Model:

Reuben Hill *et al.* described the first comprehensive model of family stress in 1958, named the ABCX model. It became the cornerstone of the family stress field of knowledge and as such 'remained virtually unchanged for 30 years.' (Walker, 1985) It is represented graphically in Figure 4, below. The data used in the formulation of this model came from the adjustment of Iowa families to the crisis of war separation and reunion (Patterson and Garwick, 1994).

It focuses on four factors which were identified by the researchers as key to the development of family stress (Gauthier-Weber, 2011):

- A: The crisis precipitating event. This represents both the event and the associated difficulties it causes. Hill further clarified this by describing the event itself in his research as 'a situation for which the family has had little or no preparation for and

must therefore be viewed as problematic'. In the context of this study, an example would be the diagnosis of paediatric epilepsy.

- B: The 'crisis-meeting resources'. Hill described this factor as being made up of features within the organisation of the family that are protective in preventing the escalation of an event into crisis. These could include parents' employment or level of education in addition to support systems from the family's community.
- C: The family's perception of the event. This factor is used to represent how a family view and define the event in relation to themselves.
- X: The crisis itself. It is formed by the combination of factors A, B and C which result in the extent of crisis.

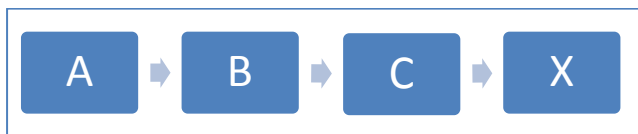


Figure 4: The ABCX Model

The majority of Hill's studies focused on factors B and C. He proposed with regard to these, that the positive or negative perception of the stressor by the family has a profound impact on the family's capability to adequately adapt and consequently, the final outcome of the crisis (Walker, 1985).

In addition to the primary framework, Hill's research also suggested several systems of classifying the event in order to help predict family response. First, he categorised the event by whether it was an internal stressor, *i.e.* caused by a family member or an external event, as was the case in his study, the effect of war. Second, a separate classification method was designed based on the type of event. Hill offered four options within this system (Walker, 1985):

1. Loss of a family member
2. Unexpected addition of a member
3. Demoralisation or loss of family unity
4. A combination of the above.

This archetypal work has been the basis of many later theories within the realm of family stress. Its main strengths lie in the primary identification of the key variables, which were previously un-described. In addition, it comprises a logical structure with semantic clarity. Furthermore, later research has also demonstrated the content and construct validity of this model (Dong, 2011).

However, as the knowledge base has expanded following this original construct, several limitations have been identified. Firstly, the definitions of the variables make the model difficult

to apply, as they label each factor as a discrete variable instead of as a continuing process. A further issue related to this, is that research suggests that chronic strains have a greater impact than single life events on their own (Gauthier-Weber, 2011). By failing to recognise the ongoing nature of a stressor, a certain amount of bias may be introduced into the model.

Another difficulty arises in separating the event from the crisis, as these may often be interconnected. Furthermore, although internal and external classification of the event can be seen to affect response, the other subdivision regarding the type of event is not broad enough so that not all events will be possible to classify. With regard to type of event, it is difficult to see the value it offers in terms of understanding family responses.

Another potential bias of the ABCX lies in the type of family used to define this framework. Using military families allowed for consistency in terms of exposure to the stressor being measured, however, it is important to note that this type of family, due to profession related constraints, may differ greatly from non-military associated families. As such, generalisation to the wider population could result in some inaccuracies.

The recognition of these potential shortcomings led to several proposed adjustments to improve the original construct. The most notable of which, was by Burr *et al.*, in 1970 (McCubbin *et al.*, 1980). Using the initial model, they then inferred 6 variables for family response to stressors and an additional 12 factors specifically for a family's response to a crisis situation (McCubbin *et al.*, 1980). The most important of the variables that they identified were: family vulnerability, which intimates the susceptibility of a family to be overwhelmed by a stressor/ stressors and family regenerative powers, which represent each family's capacity to recover from the consequences of a crisis and resume normalcy (McCubbin *et al.*, 1980).

1.3.2 The Double ABCX Model:

In 1982, McCubbin and Patterson presented the Double ABCX model, illustrated in Figure 5. Based upon the classical design put forward by Hill, it builds on the existing theory by acknowledging that family stress does not end with the production of a crisis. It reflects on this idea by expanding the ABCX model to include a pre- and post- crisis time period. This was done by introducing a further four variables. These were added to model the consequences experienced by the family in the aftermath of the crisis and the associated coping requirements (Lavee, 1985):

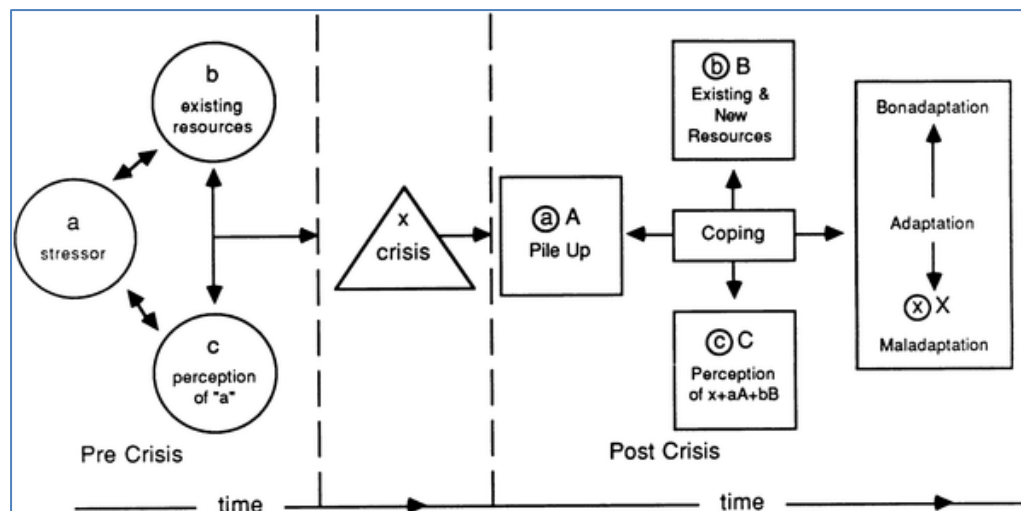


Figure 5: The Double ABCX Model (Gaudet, 1989)

- aA: refers to a cumulative 'pile-up' of demands or additional stressors occurring either in the pre or post crisis period (McCubbin et al., 1980). Examples of these stressors include: required role changes, previous unresolved family strains *etc.*
- bB: signifies the adaptive resources of the family. These may be pre-existing or developed in response to the stressors. It encompasses personal resources such as self esteem, familial ones such as good cohesion and communication between members but also social support from external institutions (Lavee, 1985, McCubbin et al., 1980). The relevance of this factor (within the model) lies in the relationship it has between the demands and the ultimate adaptation of the family.
- cC: represents the coherence and perception of the family unit. It describes how the family assesses the crisis situation and its associated demands in relation to themselves as a whole (Lavee, 1985). Within this, the authors also highlighted several additional factors that influencing coherence such as: family integrity and previous experience (McCubbin et al., 1980).
- xX: reflects the outcome of the family's progression and adaptation in response to the crisis. Here, adaptation is described as a continuous spectrum, with the two ends: 1) Maladaptation: this indicates a disparity between the demands enforced by the stressors and the family's ability to adapt/respond to them. It is more likely to be associated with a negative outcome. 2) Bonadaptation: represents the opposite side of the continuum (Lavee, 1985). It suggests that the family in question have maintained a minimal difference between factors aA and bB and therefore remain in equilibrium.

Although linear in its progression, it does however, greatly improve upon the ABCX limitations by allowing for the fact that a crisis may be ongoing, over a period of time, rather than solely an acute event. Another improvement is the recognition of the effect that other previous,

stressors (represented by factor aA) may have on the degree of stress experienced by the family.

Furthermore it also recognises that not all stress leads to a crisis. As illustrated above by Figure 5, existing resources and appropriate coping mechanisms may allow the family to adapt such that they are able to resist the impact provoked by the stressor.

Some drawbacks, however, do still exist. Both the ABCX and the Double ABCX require an event to initiate stress. Therefore they make the assumption that prior to the disruption provoked by the stressor, behavioural and social patterns within the family exist in a homeostatic state.

1.3.3 The Family Adjustment and Adaptation Response (FAAR) Model

The FAAR model, developed in 1983, incorporates several elements of its contemporaries. As a bio-psychosocial model it aims specifically to deal with family adaptations in relation to health matters (Patterson, 1988). It is designed around three core systems: the individual, the family and the community as illustrated in Figure 6. Around these, key associated factors are linked. By organising these core elements into a hierarchical system, it helps resolve a recurrent issue of family stress: how stress is related or transferred between individuals within the same unit (Patterson, 1988, Boss and Greenberg, 1984)

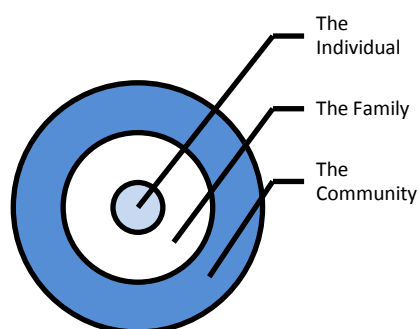


Figure 6: Representation of the FAAR Model Hierarchy of Systems

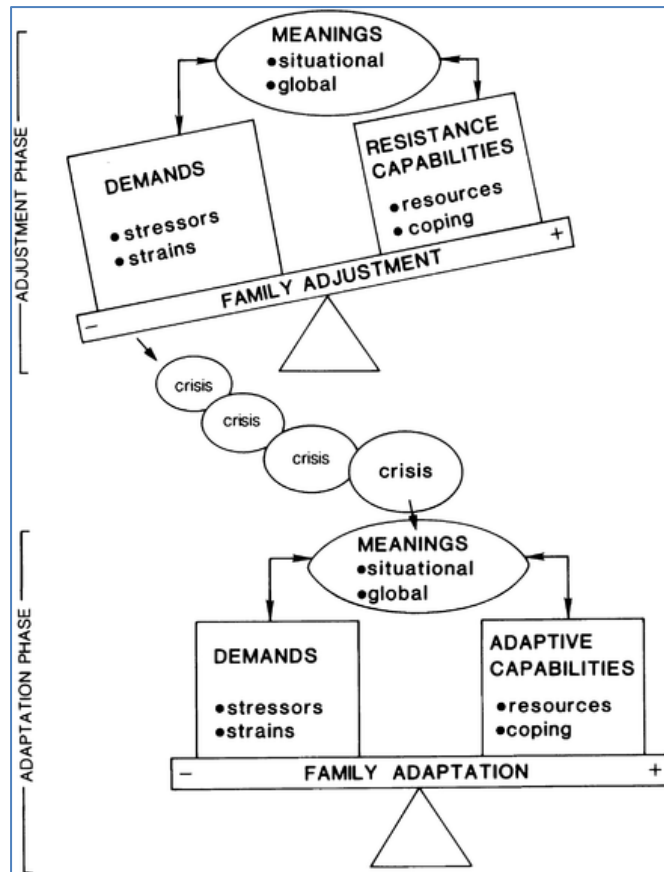


Figure 7: The FAAR Model (Patterson, 1988)

For each of the three different systems, adaptation and adjustment factors are both considered separately using a framework (illustrated by Figure 7) before being combined together at the end as an outcome. As in the double ABCX model, it accounts for pre- and post- crisis variables whilst also incorporating an element of the transactional model via the community subheading. Although inclusive in terms of all the factors associated with this model, this can also arguably be its main drawback. The complexity and different dimensions of this theoretical framework make it harder to apply to actual situations.

1.3.4 The Circumplex Model of Marital and Family Systems

This model, published *circa* 1989, differs from the existing models by way of its formulation. Rather than to illustrate a particular theory, it was specifically developed for use in 'clinical assessment, treatment planning and outcome effectiveness in research.'

Three key domains were chosen as the focus of this model based on their perceived relevance to family systems. They are represented in a circular, overlapping format as described in Figure 8. These domains are:

- Flexibility/Adaptability: Defined by the model as the amount of change in leadership, relationship roles and relationship rules
- Family Cohesion: Refers to the levels of emotional bonding that family members experience towards one another. Within this, several specifics have been highlighted, such as boundaries, decision-making and dependency. When assessing this focus with regard to outcomes it is important to note the way in which cohesiveness is scored. The spectrum, as outlined by Olson ranges from low levels (or disengaged) to high levels (enmeshed)(Olson, 2011). It is believed that for optimum stress management a balance between the two poles is most desirable and predictive of better outcomes.
- Communication: This last variable is critical. The communication within the family be it verbal/nonverbal/etc has been highlighted as a critical factor in facilitating movement between the other 2 dimensions.

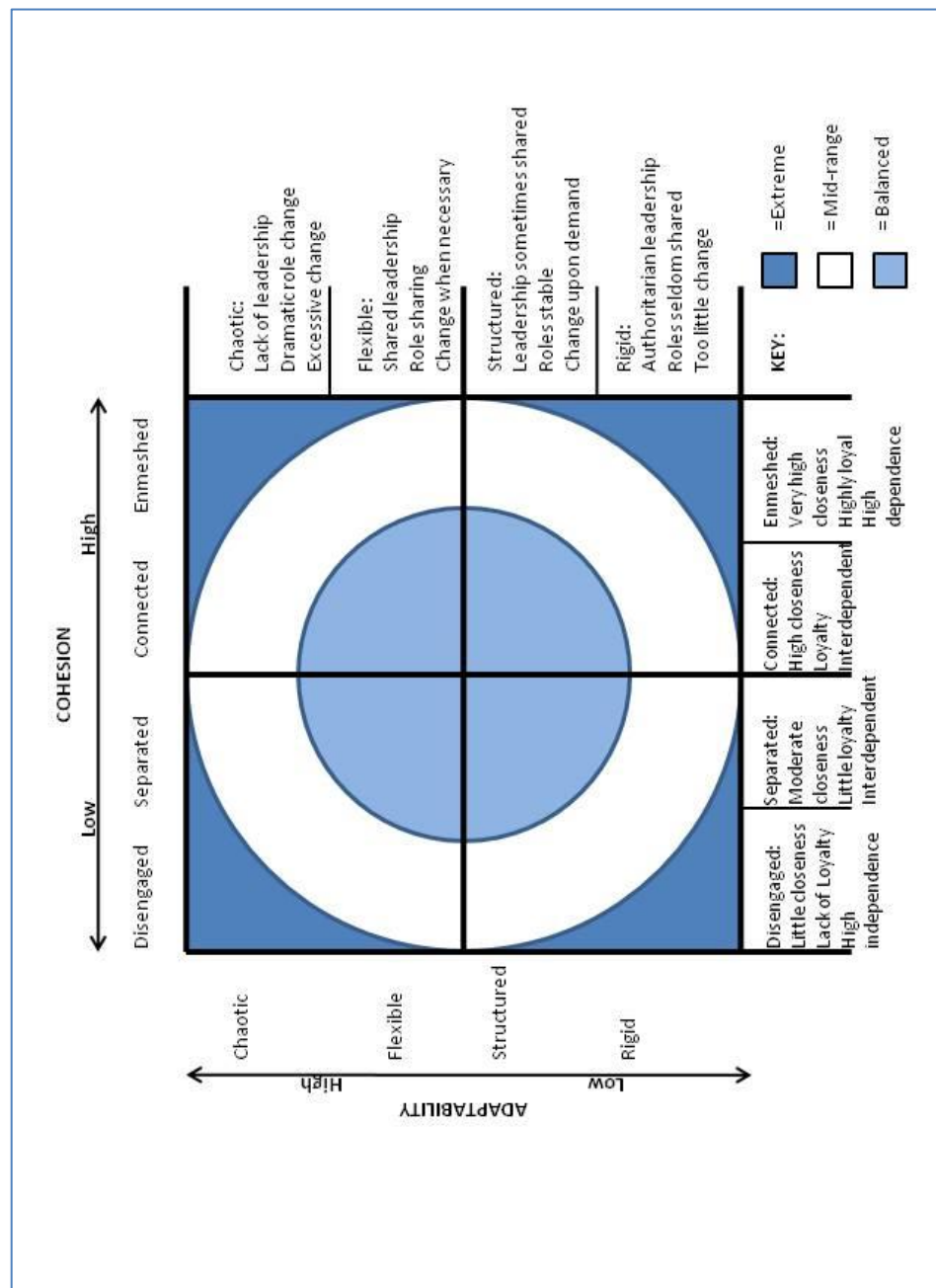


Figure 8: Diagram of the Circumplex Model of Family Systems. Adapted from (Olson et al., 1979, Olson, 1988)

1.3.5 The Family Resiliency Model

The Family Resiliency Model is one of the most recent in family stress theory, first developed by McCubbin *et al.* in 1991 and which underwent a series of changes up until 1996. As suggested by its title, the main emphasis is on understanding why some families are more 'resilient', both to crises and their associated recovery, whereas others appear to be more susceptible to stressors.

Resiliency is fast becoming a popular term within the area of family science and as such has been defined slightly differently by separate authors: McCubbin first described it as 'characteristics, dimensions and properties of families which help them to be resistant to disruption in the face of change and adaptive in the face of crisis situations' (Hawley and DeHaan, 1996). Whereas a more recent definition, as used by the USDA Family Resiliency Network as 'the family's capacity to cultivate strengths to positively meet the challenges of life' (Hawley and DeHaan, 1996)

The framework itself begins in the conventional manner, by starting with a stressor. Following on from this, a response is based upon the following factors: (V) which represents family vulnerability, (A) the pile-up of existing stressors and tension and (T) the functioning patterns within the family (Hall et al., 2012). Thus far there is significant overlap with existing constructs, namely the Double ABCX. These three factors then interact with each other along with (B) the family resistance resources. Examples of these may include: family communication, community supports *etc.* Next in this construct follows (C) another familiar variable: the family's definition of the event. Following on from this comes (PSC) *i.e.* the family's individual problem solving and coping strategies. With the end result of the model being (X) bonadaptation, maladaptation or crisis (Hall et al., 2012).

Overall, although this model introduces some novel ideas, there is significant overlap with existing family stress constructs. The main contribution of this model is the recognition that each family has unique traits that may affect the way in which it responds to stressors and that this response is not solely defined by the resources and skills they have at their disposal. Additionally, the ongoing appraisal aspect of this model, helps combat the over-riding linearity weakness of previous family stress models. The main critique however of the resiliency model is its perceived lack of clarity. Definitions of different component factors overlap, giving the impression of redundancy. This is the case particularly the repetition of factor (T) and its similarity with (TT).

1.3.6 The Transactional Model

Despite being initially modelled for individual stress, the transactional model of coping, has been shown to have much relevance when applied to a family dynamic (Oliveri, 1980). It was first put forward in 1977 by Lazarus and Folkman, two American psychologists considered pioneers within the realm of social and family sciences (Oliveri, 1980). While it does share some features previously highlighted by the ABCX model, it also introduces several novel factors and mechanisms (discussed below).

The core assumption of the transactional model is that a stressful experience is construed as a series of personal-environmental transactions, illustrated in Figure 9 (Twente, 2010).

The 'stress process' as outlined by this model, is initiated by the presence of an external environmental stressor. This leads to the primary appraisal, which refers to the individual/family's first evaluation of the stressor with regard to its severity, relevance, ability to be controlled *etc.* Following this, is the subsequent secondary appraisal occurs: the analysis of coping resources and available choices with which to address the stressor.

The assessments made at each of the appraisals may mediate the impact of the original stressor on the outcome. Adaptation and outcome are difficult concepts to infer in a psychological construct such as this. However, with regard to the transactional model, the factors concerned in evaluating outcome are clearly defined and include aspects such as: functional status, emotional well-being, familial relationships and (in relation to medical stressors) health behaviours.

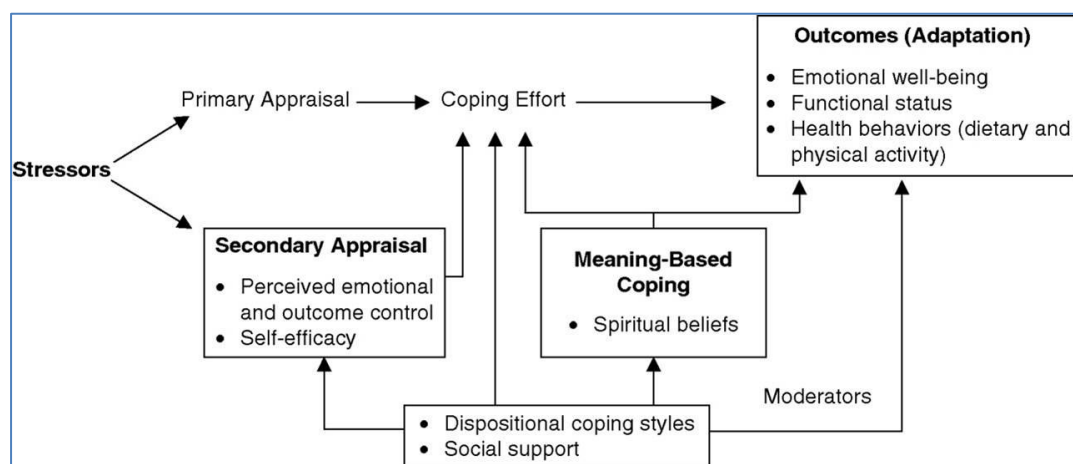


Figure 9: Transactional Model (Samuel-Hodge et al., 2008)

When determining the utility of the transactional model with regard to family stress theory, its key input has been in the introduction of an interactive element between the individuals and the stressor. This is in contrast to the ABCX based models, where the outside events impact the individual and not the reverse. It also marks the first family stress model to specifically recognise the input of spiritual and cultural beliefs which while indirectly acknowledged in other works, were not considered separately in their own right.

A further novel concept outlined by the transactional model is that of secondary appraisal, which in psychological context may also be termed habituation. It refers to the simple learning process by which repeated exposure to a stimulus results in a decrease in the previously elicited behaviour. In this way, the model proposes that people can be 'taught' to cope with stressors and therefore manage their stress. By learning how to frame or perceive events which may be considered stressors and identify effective intervention factors it may be possible for individuals or families to minimise the levels of stress that they experience.

Also, in contrast to the previous models, it is not an event driven framework. This has the benefit of allowing greater flexibility and making it easier to apply to real-life scenarios.

However, this model also has several weaknesses. The dependence of individual cognitive processes in progressing along the framework makes it more difficult to apply to a family oriented scenario (Smith, 1984). Furthermore, the model does not directly account existing or continuing strains into the equation. Rather each stressor requires a separate pathway through the framework.

1.3.7 Summary of the Theories of Family Stress

Table 1: An overall comparison of the key models of family stress theory

Family Stress Model	Summary of the Key Concepts	Strengths	Weaknesses
The ABCX Model, 1958	<ul style="list-style-type: none"> Stress is a process caused by a series of events and resulting in crisis' 	<ul style="list-style-type: none"> First to define & outline key concepts. 	<ul style="list-style-type: none"> Linearity No recognition of the period after onset of stress.
The Double ABCX Model, 1982	<ul style="list-style-type: none"> An evolution of the ABCX model and first to describe the presence of a post-crisis period. 	<ul style="list-style-type: none"> Recognition of long-term as well as acute stressors Acknowledges potential for multiple stressors and their cumulative effect (variable aA) 	<ul style="list-style-type: none"> Linearity Assumption that the presence of a stressor automatically results in a crisis
The FAAR Model, 1987	<ul style="list-style-type: none"> Focuses upon family adaptation and adjustment and describes them as a cyclical process over time. 	<ul style="list-style-type: none"> Continuous, cyclical process Highlights and separates adaptation and adjustment 	<ul style="list-style-type: none"> Complex Difficult to apply Does not directly specify what is included within the family resources factor
The Circumplex Model 1989	<ul style="list-style-type: none"> Hypothesises that balanced levels of cohesion and flexibility lead to healthy family functioning 	<ul style="list-style-type: none"> Useful for application in clinical and research purposes 	<ul style="list-style-type: none"> Perceived over simplification of factors: e.g. communications viewed as positive or negative.
The Family Resiliency Model, 1991	<ul style="list-style-type: none"> Each family's resources & attributes helps determine their susceptibility to stress. 	<ul style="list-style-type: none"> Considers the individuality of every family members as well as the unit as a whole 	<ul style="list-style-type: none"> The extremely detailed and complex theoretical nature of the model may limit its practical applications Element of redundancy in factors (e.g. T and TT)
[The Transactional Model, 1977]	<ul style="list-style-type: none"> Stress is a product of the relationship between personal and environmental transactions. 	<ul style="list-style-type: none"> Incorporates coping and habituation Outcomes specifically mention health related. 	<ul style="list-style-type: none"> Does not acknowledge presence of existing or continuous stressors

Chapter 2: Paediatric Epilepsy

This chapter seeks to highlight and discuss the key facts of paediatric epilepsy from its epidemiology and aetiology to the clinical features, management and long-term outcomes. This knowledge is crucial to the thesis in order to gain an understanding of the way in which a new diagnosis of paediatric epilepsy may impact a family. Furthermore, it allows a greater insight and appreciation for the variety of potential factors which may influence stress levels within an affected family during such a time.

2.1 History and Definitions:

Epilepsy is one of the oldest documented human illnesses (Diamantis et al., 2010). The term epilepsy derives from the Greek verb '*epilambanien*' meaning to seize or possess (Magiorkinis et al., 2010). This complex and chronic condition was first medically described by the ancient Babylonians in approximately 1000BC and the ancient Greeks who named it 'the sacred disease' due to their belief that it was a spiritual illness caused by the gods (Milton, 2010). It was not until the 6th century AD, that the lauded Greek philosopher Hippocrates rejected the divine nature of the illness and was the first to postulate that epilepsy was actually a condition affecting the brain (Magiorkinis et al., 2010). Following on from this, the understanding of epilepsy has greatly progressed over the ages.

Epilepsy is today described as 'a neurological disorder in which sudden and excessive paroxysmal discharges of cortical neurones within the cerebral hemispheres lead to disturbance of function, such as motor, sensation, consciousness or perception' (Berg and Scheffer, 2011). These paroxysmal events, characteristic of epilepsy are commonly known as seizures. These in turn are defined, as per the International League Against Epilepsy (ILAE) guidelines as: 'a transient occurrence of signs and/or symptoms due to abnormal excessive or synchronous neuronal activity in the brain' (Berg et al., 2010).

2.2 Epidemiology

On a global scale, recent data from the World Health Organisation (WHO) suggests that approximately 50 million people are affected by epilepsy (WHO, 2005) and that worldwide cost of epilepsy is thought to represent 1% of the total global illness burden (WHO, 2005).

However, although present across all nations, the geographic distribution of epilepsy is not evenly spread, as seen in Figure 10. For reasons as yet unknown, greater prevalence is found in more developed countries and significantly lower rates in areas such as South-East Asia (WHO, 2005). However a potential reason behind this may be down to under-diagnosis.

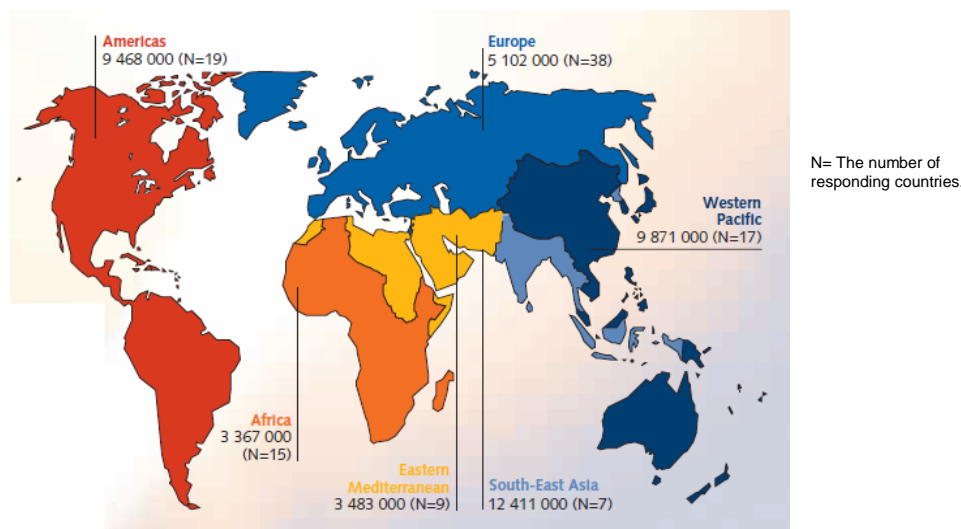


Figure 10: Number of People with Epilepsy per WHO Defined Region (WHO, 2005)

In England alone it is estimated that between 362,000 and 415,000 people are affected by epilepsy. Annual incidence rate in the UK is in the region of 50 people per 100,000, with the prevalence of active epilepsy estimated at 5-10 cases per 1,000. As a result of this, the cost of epilepsy (both direct and indirect) to the British economy is approximated as £2 billion per year (NICE, 2012b).

In contrast to many other conditions, age distribution in epilepsy doesn't follow a classical Gaussian distribution (Sander, 2003). Meta-analysis of descriptive epilepsy data from developed countries indicates that prevalence of epilepsy peaks first during childhood and adolescence remains stable throughout adulthood and then gradually increases again with age after 50 (Banerjee et al., 2009). In addition, this analysis indicated that although some smaller-scale studies found a higher male to female prevalence ratio, there was in fact no significant absolute difference (Banerjee et al., 2009).

2.3 Aetiology

The aetiology is still not fully understood as a multitude of different factors have been implicated. However as per ILAE recommendations the following broad categories may be used to help segregate the likely causes behind epilepsy:

1. Structural-metabolic causes. An example of which is cerebral palsy
2. Genetic causes. These include conditions such as tuberous sclerosis and La For's disease.

3. Unknown causes. Despite huge advances in medical investigations and radio-imaging, in as many as 70% of all cases, the cause for the development of epilepsy remains unidentified (Baker, 2011).

The onset of epilepsy occurs predominantly in childhood and in fact 70% of people who go on to develop epilepsy do so within the first 2 decades of life (SIGN, 2005). But, it should be noted that epilepsy does also first present in adults, albeit with a few differences: their symptomology is considered more homogenous and it is more likely to occur secondary to another condition, such as stroke, brain tumour or trauma (Sander et al., 1990). This is primarily because the brain continues to mature through childhood and adolescence, therefore as a result of the changing neurobiology epilepsy in children changes and evolves during their development (Holmes and Ben-Ari, 2001). However, although the distinction is important, the focus of this research and subsequent thesis is paediatric epilepsy, therefore the remainder of this chapter will be centred upon this.

2.4 Clinical Features and Classification

The principal clinical feature in epilepsy is, by nature, the seizures themselves. However, there is huge heterogeneity in seizure characteristics, for example: frequency, duration, associated automatisms, the presence of tonic/tonic among others.

Most seizures are relatively short-lived and last a few seconds or minutes. However, in some cases, status epilepticus occurs. This is defined as a prolonged seizure lasting greater than 30 minutes, or a cluster of seizures in which the person does not fully regain consciousness between each one. It conveys considerable risk to the affected individual. It is estimated 75% of all status epilepticus cases are first presentations of paediatric epilepsy (SIGN, 2005).

Classification within paediatric epilepsy is currently the subject of much debate. Historically, if classified, it was based solely upon clinicians' observations and opinions (Panayiotopoulos, 2012). More recently, a variety of different methods of subdivision have been employed such as: seizure type, EEG results, syndromic classification *etc* (Mindruta et al., 2011).

However, in 2010 the ILAE, a key governing body regarding epilepsy worldwide, published a global report followings its recently revised terminology and classifications. This has seemingly had a positive effect by giving clarification and a globally applicable system of categorisation in epilepsy. One of the principal points raised was distinguishing between generalised and focal epilepsies *i.e.* whether the seizure involves both cerebral hemispheres and is therefore generalised or whether there is an identified seizure locus in which case the epilepsy is deemed focal. This is illustrated below in table 2.

Accurate classifications within epilepsy have many important practical implications, primarily for determining appropriate management regimens. Different types of seizure have been documented to respond better to certain medications and indeed certain antiepileptic drugs (AEDs) are known to worsen certain epilepsy syndromes (Appleton, 2009b). The classification, Table 2, may also be useful in providing individuals and families with the most relevant information for their specific condition. This could in turn influence the levels of stress experienced by families.

Table 2: ILAE Schematic of Classifications in Paediatric Epilepsy, Adapted from Berg *et al.* (Berg et al., 2010)

Epilepsy classification	Sub groups
Generalised	Tonic-clonic Absence: <ul style="list-style-type: none"> • Typical • Atypical • Absence with special features Myoclonic: <ul style="list-style-type: none"> • Myoclonic • Myoclonic tonic • Myoclonic atonic Clonic Tonic Atonic
Focal	Impairment of consciousness: <ul style="list-style-type: none"> • Dyscognitive • Evolution to bilateral convulsive seizure No impairment of consciousness: <ul style="list-style-type: none"> • Motor and/or autonomic components • Sensory and/or psychic components
Unknown	Epileptic spasms Other

2.5 Diagnosis and Investigations

There are no exact answers for the diagnosis of paediatric epilepsy, although the respective National Institute for Clinical Excellence (NICE) and Scottish Intercollegiate Guideline Network (SIGN) bodies have published some evidence-based guidance for use in the U.K.

The conclusions are most often clinical and centred on the patient and/or carer's history. However a variety of investigations are available to help confirm the diagnosis. A group of leading experts have proposed that diagnosis of paediatric epilepsy should be decided based upon the consideration of 4 key areas (Appleton, 2004):

1. Seizure type
2. Epilepsy syndrome
3. Aetiology
4. Presence of co-morbidities

These specifications are deemed important because an estimated 124,500 people in the UK alone, have been given an incorrect diagnosis of epilepsy (NICE, 2012b) and are therefore taking unnecessary medications. In addition, several other conditions in children may present in a manner similar to epilepsy. Therefore care must be taken to accurately identify the cause in order to ensure the best outcome for the patient. Examples of such differential diagnoses are described Table 3, categorised by child age.

Table 3: Differential diagnoses for paediatric seizures categorised by age. Adapted from NICE guidance (NICE, 2012a)

Differential Diagnoses		
Infant	Toddler	Older Child
Cardiac arrhythmias	Cardiac arrhythmias	Cardiac arrhythmias
Hyperekplexia	Reflex anoxic seizure	Neuro-cardiogenic syncope
Structural cardiac lesion	Cyanotic Breath holding attacks	Reflex anoxic seizure
Benign myoclonus of infancy	Hyperekplexia	Hyperekplexia
Paroxysmal dystonia	Myoclonus	Myoclonus
GORD/Sandifers Syndrome	Paroxysmal dystonias	Tics
Benign paroxysmal torticollis	Sandifers Syndrome	Paroxysmal dystonias
Alternating hemiplegia	Benign paroxysmal vertigo/torticollis	Benign paroxysmal vertigo/torticollis
Infantile spasms	Migraine	Migraine
Self-gratification behaviour	Cataplexy	Eye movement disorders
Shuddering attacks	Akinetic (drop) attacks	Episodic ataxia
Sleep myoclonus	Overflow movements	Cataplexy
	Self-gratification behaviour	Atonic (drop) attacks
	Stereotypies/ritualistic behaviour	Daydreams
	Head-banging	Hyperventilation panic/anxiety disorders
	Confusional arousal	Non-epileptic attack disorder
	Night terrors	Pseudo or psychogenic seizures
		Stereotypies/ritualistic behaviour
		Confusional arousal
		REM sleep disorders
		Night terrors

When investigating epilepsy, the suggested first line investigation which is routinely performed is an electroencephalogram (EEG) (NICE, 2012d). This involves placing 12 electrodes on the scalp which allows the monitor to read electric conduction through the brain. In addition to observing the electric waveforms at rest, photic stimulation and hyperventilation, which are known triggers for some forms of epilepsy are also carried out as part of a standard EEG to improve the likelihood of visualising a seizure on the EEG recording(NICE, 2012d). The

resulting scan can then be examined to look for any electrical waveform patterns consistent with epilepsy.

A normal EEG alone however, does not exclude a diagnosis of epilepsy. If deemed necessary, further and more complex EEG investigations may be carried out, such as a sleep deprived EEG, since excessive tiredness is another documented trigger for epileptic seizures. Alternatively, an ambulatory EEG may be carried out. This involves keeping the electrodes on for a longer period of time, usually 24 hours. The patient is able to carry out their normal daily activities but the longer duration of the investigation makes it more likely for the device to pick up a suspected seizure.

If the EEG investigations have not yielded any results and there are still queries as to the nature of the seizure-like event, other options available include various forms of imaging such as magnetic resonance imaging (MRI) or computed tomography (CT) scans. These are particularly useful in excluding any structural abnormalities in the brain which may be causing the seizures. However, because these investigations are seen as more invasive (often requiring the child to be anaesthetised beforehand) NICE guidelines recommend that, barring special considerations, MRI should only be undertaken if (NICE, 2012d):

- The development of epilepsy occurs before the age of 2 years or in adulthood
- There is any suggestion of a focal onset: either from the history, examination or EEG findings (unless there is clear evidence of benign focal epilepsy)
- The child's seizures continue, in spite of first-line medication

The indications for CT scanning are confined to: a contra-indication to MRI or for use in an acute setting to determine whether a seizure had resulted from an acute neurological illness or lesion (NICE, 2012d).

Alongside the relevant neuro-imaging, if there is still a cause for concern, or the need to exclude other possible causes, additional investigations may also be carried out 'at the discretion of the specialist(NICE, 2012d)'. Examples of these include: an electrocardiogram, urine and blood biochemistry, such as plasma electrolytes, blood calcium/glucose levels.

In addition to physical investigations, another important avenue to explore in children is a neuropsychology assessment (NICE, 2012d). This enables the evaluation of any associated cognitive dysfunction or learning difficulties which may also impact the child's quality of life.

2.6 Management

Management is focused on symptom control because, as yet, there is no cure for epilepsy, although it is important to recognise that not all epilepsies require intervention. The decision to start treatment is usually based upon a risk-benefit analysis: whether the child's quality of life is affected significantly by the seizure and post-ictal events enough that it outweighs the potential negative sequelae that may be incurred by commencing therapy. It has been further suggested that treatment should only be started following a confirmed diagnosis of epilepsy and after a minimum of 2 seizures (NICE, 2012c). This response stems, in part, from statistics indicating that in children who have a first unprovoked generalised seizure with an otherwise normal medical and developmental history, only 30-40% will go on to have further seizures (Shinnar et al., 2000).

The mainstays of treatment are AEDs. These may be broadly divided into either preventative or acute management. Preventative medications are taken daily in order to reduce or completely stop the development of seizures. NICE and other regulatory bodies all recommend monotherapy with regard to these medications. Should side-effects occur with the first-line option, monotherapy should be sought with a second-line agent (with care being taken during the cross-over period between the two)(NICE, 2012c).

Acute acting or 'rescue' medications are often prescribed alongside preventative treatments for use during prolonged seizures or status epilepticus. Buccal midazolam or rectal diazepam are the most commonly used and advice is to administer these if a seizure lasts for more than 5 minutes (Klimach, 2009). An explanation for this practice is that although short-lived seizures are almost always harmless to the child, a conservative estimate of 5 minutes is given to the time after which neuronal damage may occur (Hoch, 2010, Auer and Siesjo, 1988).

AEDs often work extremely effectively in managing epilepsy, with studies showing that up to 65% of patients respond well (based on an adult and child sample population) (Appleton, 2009b). However, they are associated with a myriad of side effects. The most common of these are Type A drug reactions i.e. dose-dependent, predictable effects including: sedation, lethargy, weight gain and behavioural problems (Appleton, 2009b). Less frequent, but often severe are Type B, idiosyncratic reactions, effects such as: hypersensitivity reactions (Appleton, 2009b).

When treating adolescent girls (and women of child bearing age) discussion of AED side effects and drug interactions is of particular importance as the majority of this drug class are known teratogens and due to their interaction with P450 enzymes, they are additionally known to alter the efficacy of hormonal contraception methods (Crawford, 2005) .

Along with medication, patient and parental education is also a key part of management. Understanding of their condition and how it may impact areas of their lives is important for the individual but also the family. Education of what to do for someone when they are having a seizure is also key, as well as ensuring that the child's teachers and (depending on age) friends are also aware of this. In some cases psychological treatment for both the child and carers may be of assistance.

Unfortunately, not all cases of childhood epilepsy are easily treated; approximately 30% of paediatric patients suffer with intractable epilepsy *i.e.* when greater than two different AEDs have been tried but had little or no effect (McTague and Appleton, 2011). In cases such as these, more invasive forms of therapy may be warranted, examples of which include: the ketogenic diet, vagus nerve stimulation and epilepsy surgery.

2.7 Prognosis

Prognosis for paediatric epilepsy is usually extremely good. The overwhelming majority continue in mainstream schools and go on to lead normal lives. There is, however, a moderately increased morbidity and mortality associated with the condition and its management (Nikolas et al., 2007). The standardised mortality ratio in the UK for people with epilepsy has been estimated as 2.1 when compared to the general population (Berg, 2001).

In some cases, paediatric epilepsy is associated with other co-morbidities. A large scale study in the USA estimated that these were apparent 26% of cases (Baca et al., 2011). These may influence the prognosis and quality of life of the child. The most common of these are psychiatric and neuro-developmental conditions including: attention disorders, learning difficulties, features of autistic spectrum disorder and mood disorders among others (Baca et al., 2011). The Isle of Wight epidemiological studies indicated that 28.6% of children with 'uncomplicated' epilepsy experienced psychiatric disturbances compared to 6.6% of those children without (Rutter et al., 1976). More severe issues are less common and usually associated with a specific epilepsy syndrome, such as profound developmental delay in children with Dravet's syndrome (Camfield et al.).

Another aspect when considering the long term prognosis is the possibility of Sudden Unexpected Death in Epilepsy (SUDEP). A well accepted definition, as proposed by (Nashef et al., 2012) states it as: *'the sudden unexpected, witnessed or un-witnessed, non-traumatic and non-drowning death of patients with epilepsy with or without evidence of a seizure, excluding documented status epilepticus and in whom post-mortem examination does not reveal a structural or toxicological cause for death.* Although this is a relatively rare occurrence, a recent population cohort study which followed children with epilepsy over a

period 40 years, found that SUDEP occurred in 9% of cases and accounted for 38% of all deaths (Devinsky, 2011). Whilst the onset of epilepsy developed in childhood, it is worth clarifying that the majority of SUDEP cases did occur during adulthood. This is in concordance with previous findings indicating that it is less common under the age of 14 than in adulthood (Devinsky, 2011). However, it still is an important consideration and therefore it is now required for the treating physician, upon giving a new diagnosis of paediatric epilepsy to explain and discuss this with the parents, along with providing the appropriate advice on risk factors and minimising the likelihood of SUDEP.

2.8 Social Issues

In addition to the clinical implications of living with epilepsy, this condition also raises several considerations regarding social involvement. An important part of this is based upon recognising potential risks that epilepsy may impose onto a child through their interactions with society. Therefore, the majority of suggested limitations are centred upon maximising safety.

An obvious issue is the communication and involvement of the child's school, who should be notified following a confirmed diagnosis of epilepsy (Baker, 2011). Steps should be taken to ensure that relevant staff members are suitably aware: firstly, on adequate supervision during certain classes (e.g. Sports and physical education) but secondly, on the appropriate management of the child during a seizure.

Some leisure pursuits can also be affected by a diagnosis of epilepsy. Activities such as, cycling, running and especially swimming should be supervised. National clinical guidelines even suggest that doors should not be locked on bathing and road crossing should always occur at designated pedestrian crossings (SIGN, 2003). Furthermore, although only a relative minority of children (estimated at 5%), with epilepsy experience photosensitivity, this can have a significant effect on their lifestyle. In childhood, it may limit them from things such as certain theme park rides, 'laser quest' attractions, discos and some video-games. On progression to adulthood, it may also impede them from aspects of nightlife, due to the high prevalence of strobe lighting (Appleton, 2009a). Interactions with their medications in combination with alcohol and recreational drugs should also be discussed.

Further considerations for older children or young adults are the restrictions placed on driving. Due to the risks posed to both the person themselves and the public as a result of a seizure whilst driving, the Driver and Vehicle Licensing Agency (DVLA) has issued strict regulations on this subject (EpilepsyAction, 2011a). In addition to driving, when considering the child's future, the topic of occupation is also relevant as an epilepsy diagnosis places some

vocational restrictions, for example: professional driving (articulated or commercial vehicles) and aeronautical control (Appleton, 2009a).

Overall, a diagnosis of paediatric epilepsy does confer some important short and long term social considerations which are unique compared to other conditions. It should be emphasised that any limitation placed on the child and their family have been put in place more to ensure their safety and that of the public rather than to actively prevent them from anything. Epilepsy Action, one of the leading independent epilepsy support groups in the U.K. stresses that, within reason, children with epilepsy should be encouraged to participate in all aspects of life; just with the appropriate support and boundaries with which they can do so safely (EpilepsyAction, 2011b).

Chapter 3: Paediatric Epilepsy as a cause for family stress

Paediatric epilepsy is unique in terms of some of the health and social implications it incurs. This chapter, therefore, aims to evaluate the different means by which paediatric epilepsy may act as a stressor in a family environment. In addition, it also aims to investigate other covariates associated with stress and its outcomes in order to fully assess the range of different family variables in play as a result of one of its members being diagnosed with paediatric epilepsy.

3.1 The Parent/Carer

3.1.1 General Paediatric Disease

Many different aspects of everyday life may serve as stressors to a particular family. However, childhood illness is a recognised and well documented cause for parental and family stress. All children are dependent upon their parents/guardians; however in the context of health concerns the parental role may assume an even greater importance. 'Parents must become care co-ordinators, medical experts, systems advocates and personal representatives' for their child, in addition to their 'regular' parenting roles (Kratz et al., 2009).

Illness or disease may be defined as 'a pathological condition causing impairment in physiological functioning characterised by an identifiable group of signs and/or symptoms (Press, 2007).' In children as in adults, they can take either an acute, chronic or relapsing-remitting course and this duration may affect the severity of the stress which they may subsequently incur.

Acute childhood illness, depending on its specific constellation of symptoms and likely outcome, may well be a stressor and exert a negative effect on the parents and family as a whole. However by the very nature of being an acute and transient occurrence there is still the expectation that this illness and consequently the resulting stress is only a temporary state and that the child will recover and the current family situation will return to its original state.

However, chronic disease, upon which the overwhelming majority of research into health-related stress has been focused, represents a longstanding presence in family life. Therefore, it often has far-reaching effects into areas such as personal and social life, family finances and behaviour. As a result, family members will have to adapt their previous roles and responsibilities in response to this potential stressor.

Many studies have shown significantly increased stress in parents of children with a chronic disease compared to control groups (Hua-Huei and Liang-Po, 2008, Plant and Sanders, 2007, Bouma and Schweitzer, 1990).

It has subsequently been demonstrated by several studies dating from the 1980s, that levels of stress and its covariates are not equal between different chronic illnesses. An example illustrating this point is the heavily cited work by Holroyd and Guthrie. This research compared levels of stress between families with a child suffering either: cystic fibrosis, neuromuscular disease or renal disease (Holroyd and Guthrie, 1986). Using the well validated Questionnaire on Resources and Stress (QRS), they found increased levels of stress between these families and their matched control groups. Within this, results showed that families from the three chronic illness groups experienced different levels of stress. In particular families in the neuromuscular disease cohort experienced the greater levels of stress, recording statistically significant higher scores than the control group on 11 of the 15 different scales ($p=0.05$). The cystic fibrosis group scored lower levels of stress than expected. Furthermore, on analysis of the different subscales included in the QRS, each illness cohort exhibited stress levels in different areas.

Although this study was able to provide statistically significant results, there were several limitations. A key factor that may have influenced the results was the small sample size ($n=16$ in each cohort) which may have distorted the findings. Furthermore the controls used for this study, although matched in terms of age, were nominated by the disease- cohort families which could be a potential cause of bias.

An additional work supporting the differences in perceived stress between medical conditions was that carried out by (Bouma and Schweitzer, 1990) which evaluated and compared stress between families which had children either with autism or cystic fibrosis. Stress in mothers was again measured using the QRS and results showed that although both groups demonstrated stress, the mothers with autistic children experienced this to a greater degree. Discussion suggested that a possible cause for this unexpected discrepancy was the difference between physiological and psychological conditions. This connection has previously been proposed in other works investigating maternal stress in relation to children with different forms of psychomotor retardation.

Based upon its clinical course, epilepsy may fall into both the chronic disease and relapsing-remitting categories. These are much less numerous than both acute and chronic conditions and therefore, perhaps unsurprisingly, have a corresponding paucity of literature on their interactions with stress. The fluctuations of relapsing-remitting diseases may impede a family's habituation and thus affect their associated stress levels. It is hoped that this study will help delineate these processes.

3.1.2 Epilepsy Specific Disease Factors

As previously described, epilepsy is a complex chronic condition, which is distinct in character from other long term neurological disorders. Therefore, though previous work regarding stress in conditions such as neuromuscular disease may guide us, it is unreasonable to assume that their findings can be directly transferable to the epilepsy population. Several papers have outlined the need for a greater understanding of the effect of family stress in paediatric illness and in particular epilepsy (Duffy, 2011, Mays, 1988). A recent study found that families and carers of adults with epilepsy report low levels of both emotional and practical support which could have far-reaching effects not only on the carer but also the patient (Rodenburg et al., 2005). This is further compounded by previous research suggesting there is a correlation between insufficient coping mechanisms on the part of the parent/carers resulting in a poor overall outcome in the child.

Other issues unique to epilepsy include a negative impact on behaviour, learning and depressive/anxiety symptoms (Stevanovic et al., 2011, Ferro et al., 2010). These are potentially important factors that it would be extremely useful to address.

A major difference between epilepsy and other clinical conditions (neurological or otherwise) is its unpredictability. This was highlighted by (Duffy, 2011) as a key factor in the levels of uncertainty and concern in parents of children with epilepsy. When adults in the USA were surveyed to identify what they felt was 'the worst thing about having epilepsy' the unpredictability of the next seizure was ranked highest with 24.7%. The important role of the unpredictable time course of epilepsy was further emphasised by (Camfield et al.) research into children with Dravet's syndrome, which is a specific and severe epilepsy syndrome characterised by frequent febrile and myoclonic seizures with associated severe developmental delay. Their work, using semi-structured interviews as their primary method, suggests that uncertainty was the primary factor influencing parental stress levels.

A further matter related specifically to epilepsy over other conditions is that of stigma. There are still widespread preconceived stereotypes about epilepsy sufferers not just in less developed countries but also in Western society (de Boer, 2010). Even in the U.K., the law allowing voidability of marriage on the grounds of epilepsy, was only repealed in 1971 (Lee, 2010). Research in adults newly diagnosed with epilepsy, carried out in the USA revealed that 24% were concerned about the reactions of others and social stigma (Morrell, 2002). Social stigma was ranked second overall (23.9%) as one of the worst things associated with having a diagnosis of epilepsy. This has a negative effect regardless of whether the stigma is perceived or enacted. This indicates that preconceived misconceptions about epilepsy on the part of the families themselves but also those around them may be a significant source of stress.

In addition to these, other documented factors associated with stress and paediatric epilepsy are those pertaining to the future of the affected child (Murray, 1993). Mothers of children with epilepsy report specific fears that their child will die during a seizure (Home and Kerirey, 1991, Wiebe et al., 2009). Management concerns are also prevalent: the effect of long-term AED therapy has also been shown to affect parental stress levels (Buelow et al., 2006, Chiou and Hsieh, 2008).

3.1.3 Covariate: Carer Health

Caregiver health is an important covariate of stress. The association between caregiving and health is modulated based upon the degree of stress that they are experiencing (Raina et al., 2004). The chronic stress associated with caring for a family member has repeatedly been shown to have a negative effect on overall health as well as on specific health domains such as, medication use, health symptoms and self-reported health.

Parents of children with epilepsy have been shown to have significantly increased prevalence of psychiatric morbidity, mainly experiencing depression and anxiety related symptomology. Several studies have documented this association, particularly among mothers (Ferro et al., 2010, Shore et al., 2002). Theories attempting to explain this association are numerous but ambiguity and uncertainty on the part of the carer combined with childhood issues such as behavioural problems have been suggested. In fact, research by (Shore et al., 2002) into carers of children with epilepsy, identified low family satisfaction, low income and child behaviour to be factors most strongly correlated with carer ill health .

These findings are particularly relevant as parental anxiety alone has been associated with a decreased quality of life for the child and the family as a whole.

3.1.4 Covariate: Carer Locus of Control

The key principle of this concept is the individual's perception of the extent to which their actions are able to impact the circumstances and environment around them (Kormanik and Rocco, 2009). Locus of control has been described as a linear scale where an individual lies on a spectrum punctuated at each extreme by high internal and high external locus:

- 1) High internal LoC: Those people who believe that situations occurring around them result primarily from their own actions and behaviours.
- 2) High external LoC: Those individuals who tend to think that events affecting them are mainly determined by outside factors, such as chance, fate, higher powers etc.

LoC is of particular interest to this work due to its relevance as a mediator between stress and well-being. This is in turn relevant to health as an individuals' locus of control has been repeatedly been shown to affect their patient outcomes:

Studies pertaining to paediatrics have shown that parental locus may have a role in determining the child's long term outcomes and quality of life scores (Barakat et al., 2005). This was demonstrated in a sample population of families where a child had sickle-cell anaemia. Children of parents with highly external loci were found to have a statistically significant decreased score on the PedsQL quality of life measure (Barakat et al., 2005). Sickle-cell anaemia shares some disease characteristics with epilepsy, namely a degree of unpredictability (sickle cell crises) on a background chronic condition. Thus a similar effect could plausibly be observed between parental/carer locus of control and paediatric epilepsy patients.

3.1.5 Covariate: Carer coping and support

As with the locus of control concept, carer coping mechanisms are thought to have a 'buffering' effect on stress and maintaining wellbeing. In addition it is increasingly recognised that coping is context-dependant i.e. that the nature of the interactions between the individual, the stressor and the environment impacts the ways in which they cope.

Work by Rodenburg in 2007 supports the negative correlation between coping and parenting stress. Based upon his sample of 91 parents of children with epilepsy, he asserts that increased levels of parental stress may be due to a general lack of parental coping resources (Rodenburg et al., 2007). This is in concordance with several other large-scale studies such as that by (Mu et al., 2005) involving 316 families and further research by (Rodenburg et al., 2011) expanding upon his previous work.

This variable is especially interesting as the healthcare system is well placed to help support parents and in many instances already seeks to provide coping resources. At Alder Hey, existing epilepsy specific resources include information leaflets ('first fit' leaflet and 'your guide to childhood epilepsy'), appointments available with the epilepsy specialist nurse, opportunities for psychology referral for both the child and/or the caregiver. Therefore this covariate lends itself to the application of interventions based upon identified needs as well as highlighting any existing areas which should be audited.

3.1.6 Covariates: Family functioning and communication

Recent work by (Rodenburg et al., 2011) emphasises that family functioning plays an important role in a child's adjustment to epilepsy and shows that there is a strong association between parental stress and family functioning. Theories suggest that carer stress may indeed act as a mediating factor between parenting variables and family functioning. It is for these reasons that family functioning and family communication may be important covariates in this study, investigating the impact on carers and family following a diagnosis of paediatric epilepsy.

3.2 Child factors

3.2.1 Covariate: Child Quality of Life

The quality of life of the child with paediatric epilepsy is also an important covariate in understanding the associated family stress from this new diagnosis.

Children with epilepsy are more likely to experience symptoms of depression and anxiety than their healthy counterparts: (Ekinici et al., 2009) reported prevalence rates of 33% with depression and 49% with anxiety issues in children with epilepsy. Psychiatric co-morbidity in paediatric epilepsy is thought to negatively impact the child's quality of life. Zero order correlation analysis of depression, anxiety and quality of life questionnaires in a sample of 60 children with confirmed epilepsy showed a statistically significant correlation between all three measures -0.39- -0.55 ($p < 0.01$) (Stevanovic et al., 2011).

In addition to childhood factors affecting quality of life of the child with epilepsy, parental anxiety has also been implicated. Research in 200 parents of children with epilepsy found statistically significant correlation between child QOL and parental anxiety measures, $r^2 = 0.50$ ($p < 0.01$) (Williams et al., 2003).

3.2.2 Covariate: Behaviour

Research indicates that children with epilepsy have higher rates of behavioural problems than those who do not, with the majority of these issues being focused around attentional disorders and internalisation (Rodenburg et al., 2011). Delineating any causal relationship between paediatric epilepsy and behavioural problems has proved complex, with some prospective data indicating that child behavioural issues may in fact precede the onset of seizures (Austin et al., 2001). However, regardless of the order in which the behavioural difficulties began it is reasonable to assume that they may still be a source of parental and family stress. In fact,

when investigating potential factors linked with behaviour, parental and family variables have been shown to be more highly associated than others, such as seizure frequency (Rodenburg et al., 2011, Austin and Caplan, 2007).

Chapter 4: Approaches to Researching Stress and Systematic Review of Relevant Measures

This chapter focuses upon the different potential methods to investigate stress and involves a detailed description and systematic review of each of the questionnaires that were chosen to be used as part of the study.

4.1 Methods of Investigating Stress

Stress may be classified as either a continuous or categorical data variable and within these categories; any raw data collected may take the form of qualitative or quantitative information.

For this study, the main interest is the ways in which stress differs between 2 different cohorts in response to family and caregiver factors and how this stress may change over time. As such, tools that allow longitudinal measures and that also have capacity to be used easily for comparative analyses are of particular use.

Previous research into stress has taken a variety of different forms. The complexity of the concept poses several difficulties to investigation, namely the interplay between the psychological and physiological responses to stress (See Figures 1 & 2) *i.e.* the degree of self-perceived psychological stress may not directly correlate to the level of change in physiological measures. As a result, both qualitative and quantitative methods have been used to analyse aspects of stress, although each have their own associated strengths and weaknesses in terms of the quality and level of information they are able to provide.

Quantitative research refers to the 'systematic empirical investigation of social phenomena via statistical, mathematical or computational techniques' (Bowling, 2009). It uses discrete and specific questions to provide numerical data. This allows the application of statistical methods in order to investigate scientific theories and hypotheses. It is frequently used within social sciences and research into stress. From its inception stress has been investigated using quantitative methods: The key link is the measurement method used; be it blood pressure readings, respiratory rates focused on physical changes or Likert scale questionnaires aiming to quantify the psychological responses to stress.

Qualitative research methods are used predominantly within the social sciences and human behaviour (Green, 2011). It employs broader questions in order to gain a more in depth understanding of the field of interest. The strength of these methods is the diversity of answers and the flexibility of data collection. However this breadth is also its key weakness in

that it makes data analysis significantly more complex and often reliant on observer interpretation or data coding practices.

Investigating the ways in which a family may be affected following the diagnosis of paediatric epilepsy and the interplay between carer stress and a host of associated covariates helps to bridge the gap between family stress theories and clinical practice. In fact some studies regarding stress in epilepsy have directly used a family stress model as a basis to their research. Examples of this include work by (Buelow et al., 2006) which used the double ABCX model as a guide to their work investigating potential factors that may represent the 'pile-up of demands' variable. Additionally work by (Pei-Fan, 2005) is one of few that used the family resiliency model in order to justify the rationale of his study design.

4.2 Literature Review of Previous Research into Stress and Paediatric Epilepsy

A search of the literature into previous studies investigating stress in epilepsy was carried out. A variety of different medical/science based search engines were used in order to obtain the maximal amount of relevant hits. Demonstrated below, in Table 4, are the search terms that were used in order to identify relevant papers. Search terms were initially kept broad in order to find the maximal amount of information. Review articles were also searched separately at the beginning before criteria were made more stringent to collect papers on more specific areas.

Table 4: Search Terms Used for Identifying Relevant Papers for Literature Review and Discussion Purposes

Search Terms	Number of Generated responses (from PubMed, Scopus, Cochrane and PsychInfo search engines)
Paediatric epilepsy + Stress	239
Paediatric Epilepsy + Family Stress	84
Paediatric epilepsy + Parent or Carer Stress	116
• Review only	24
Paediatric epilepsy + Parent or Carer Stress	
• English language	97
• Published between last 10 years	88
• Published within last 5 years	41
Parent or Carer stress +	
• Parent/Carer health	45
• Parent/Carer depression	10
• Parent/Carer coping	32
• Parent/Carer support	15
• Child quality of life	38
• Child behaviour	37

From the searches, as tabulated above, 132 potential abstracts were identified from the titles. At this stage articles were excluded if they did not appear relevant to this document e.g. papers regarding oxidative stress in paediatric epilepsy rather than person-oriented stress. The original 132 abstracts were each read then 96 were discarded if their content was not applicable to the literature review or in some cases due to the quality of the outlined methodology. The remaining 36 papers which were each read in full and evaluated using the Critical Appraisal Skills Programme (CASP). 19 of these articles were found to be of relevance to this work and were therefore discussed in varying depth within this document and referenced as appropriate. The remaining 17 were excluded after reading the full length article. This was due to methodologies used (focus was centred around survey based methods), the choice of measures used, some of which were inappropriate and the study populations used e.g. mix of adult and child samples. A graphical representation of the QUOROM statement is included in Figure 11 on the following page.

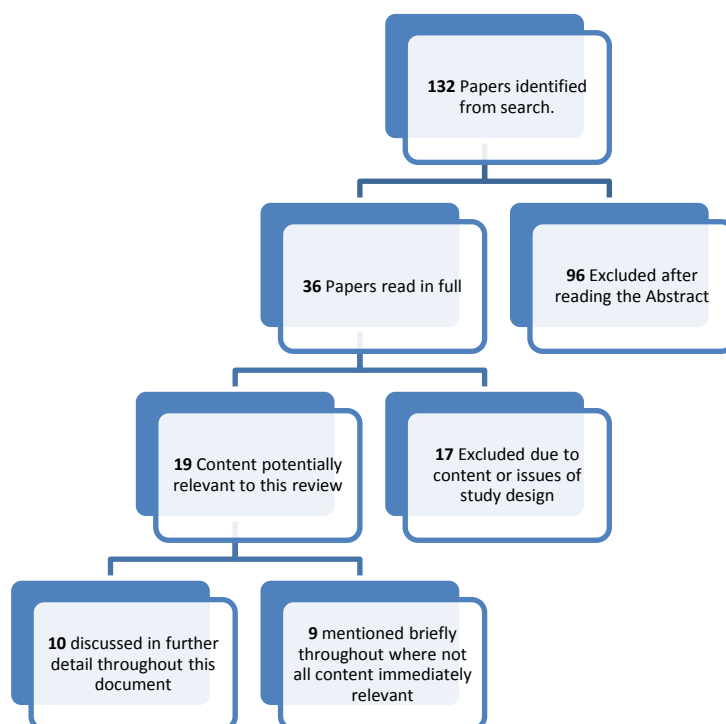


Figure 11: Graphical flow chart representation of QUOROM statement. Organised in accordance with guidance from the QUORUM conference (Moher et al., 1999)

Additionally, an example of how the CASP was used is documented below.

CASP Questions to assist with the critical appraisal of an observational study: 'The Impact of a New Paediatric Epilepsy Diagnosis on Parents: Parenting Stress and Activity Patterns' Modi *et al.* 2009 (Modi, 2009)

1 Is this study relevant to the needs of the project?

This paper is indeed relevant to the purpose of identifying existing studies of family stress and paediatric epilepsy. It is of particular interest as it is focused on new diagnosis of epilepsy, a specific area also being investigated in the research of this thesis.

2 Does the paper address a clearly focused issue?

In terms of: the population studied? The outcomes considered? Are the aims clearly stated?

Yes. The issue, as highlighted in the study title, is degrees of parental stress and activity as related to paediatric epilepsy.

The population studied is clearly identified as parents of children newly diagnosed with epilepsy. This was clarified as children diagnosed and prescribed medication (i.e. AED)

The outcomes were clearly considered as they dictated the tools used. However the article did not state what difference in the outcomes would be considered statistically significant or how that judgement would be made.

The 2 key aims of the study were clearly identified in the introduction: firstly, to compare the levels of parental stress of those with children newly diagnosed with epilepsy against matched controls and secondly to compare any difference in activity patterns between the 2 aforementioned cohorts.

3 Is the choice of study method appropriate?

The case-control design on the study is appropriate as it allowed for a control group (parents of children without epilepsy) to be included. This allows for better comparisons to be made.

Recruitment occurred at a new-onset epilepsy clinic during a follow-up visit which occurred approximately 1 month following the diagnosis of paediatric epilepsy. It was stated that this was done so as not to burden the families at time of diagnosis. In practical terms, however, one month is a reasonably long period of time with which to start adapting and coming to terms with the diagnosis.

The measures themselves were: the PSI, FSS-Seizure and daily phone diary. The PSI questionnaire (described fully on Page 43) is an extremely popular and well validated tool and therefore appropriate in nature for this study. The FSS-Seizure is a more recent tool and therefore there is less available data regarding its psychometric properties. The daily phone diary is a cued response interview in which the parent was asked to describe every activity they did in 1 day which lasted over 5 minutes and to rate their mood during it. Although this technique has previously shown useful and with high inter-rater reliability, it does pose a few limitations.

4 Is the population studied appropriate?

The population studied was the parents of children aged 2-12 years who had been diagnosed with epilepsy 1 month previously. They were recruited from a clinic in Cincinnati, USA. The population is appropriate for the study although arguably the sample size (n=59) is too small

5 Is confounding and bias considered?

Potential confounding variables were considered in terms of the demographic data collected and Fishers exact tests used to ensure that the 2 cohorts were comparable. Use of a age, gender and race-matched control group also helps limit bias.

Have all possible explanations of the results been considered?

Could selective drop-out explain the result?

Were assessors blind to the different groups?

Overall possible explanations seem to have been considered. It was not however mentioned that based upon the nature by which controls were recruited that their data may not necessarily be representative of the general population.

Information regarding attrition in the study was not presented. No mention is made of blinding; therefore it may be assumed that assessors were not blinded to the cohort allocation of the parents.

6 (Cohort study) Was the follow up long enough? Could all likely effects have taken place during the time-scale? Could the effects be transitory? Was follow up sufficiently complete? Was dose-response demonstrated?

Not applicable as this is not a cohort study.

7 Are tables/graphs adequately labelled and understandable?

Yes. All tables and graphs are clearly presented and understandable.

8 Are you happy with the author's choice and use of statistical methods, if used?

Overall I'm satisfied with the statistical analyses described. The use of Fishers exact test was appropriate given the categorical nature of some of the data. However, the choice of individual t-test for comparing the normal and control groups was an interesting choice as it makes the assumption that the data follows a normal distribution, which given the recruitment of the sample could be argued. Another potential test that could have been used is the Mann-Whitney U test as it allows for skew in the data. The use of ANOVA as part of the analysis is completely valid, given the multivariate nature of the study (and the PSI as an investigative tool)

9 What are the results of this piece of research?

Results indicated that there was no statistically significant difference in stress levels associated with parenting roles between the two cohorts. Significance was only found on the life events scale. From this sample 7% of the parents from the new onset epilepsy group were found to have elevated levels of stress.

Regarding the activity patterns no significance was found between 'generic parenting stress' and times spent in different activities. Epilepsy specific parenting stress had a moderate correlation with recreation spent within the home but no other activities.

10 Can the results be applied to the local situation?

Consider differences between local and study populations (e.g. cultural, geographical, ethical) which could affect the relevance of the study

Broadly speaking the results may be applicable to the local situation. This study was conducted in Cincinnati, USA so although also a western society it may be distinct on a cultural level, and this work reported a greater ethnic diversity than in our sample. The mean age of children participating in the work is marginally lower than that found in our sample: 6.5 compared to 7.6 years. These are all factors which could potentially affect the relevancy of this study.

11 Were all important outcomes and results considered?

Based upon this authors understanding important outcomes and results were considered. Post hoc investigations were conducted in attempt to understand significant finding on the life events scale. This led to finding that this group of carers had experienced greater life difficulties which may have had an impact on their reported stress scores.

12 Is any cost information provided?

No cost information is provided, nor were any sources of external funding noted in the acknowledgements.

For the purpose of this piece of research into stress associated with diagnosis of paediatric epilepsy, the choice of a mixed quantitative-qualitative methodology can be justified as it allows for collection of specific numerical data to be collected. This is of particular importance as it facilitates the statistical analysis of the different variables over the 2 time points at which data is collected in this study. However it also allows the addition more open ended questions to be asked. This can be considered to complement the discrete data because this study seeks to investigate stress and its correlates. It is conceivable that in the design of this study a potential correlate may have been overlooked and interaction with families in which they are able to give more detailed answers and opinions may help recognise not only weaknesses in the study but also provide ideas for the direction of work.

However, in this thesis **only quantitative data and analysis will be presented**. This is because although being sought, there is currently no funding for qualitative analysis and none of the research team have the appropriate skill set to adequately perform this work.

4.3 Questionnaires Selected for this Research

Following the decision to follow a quantitatively based, survey centred methodology, the next logical step was to appraise the questionnaires available to be used.

In order to quantify family stress as experienced by individual members, a huge range of different questionnaires have been developed within the last 2 decades alone. To help explore a variety of potential factors associated with a new diagnosis of paediatric epilepsy that may influence the levels of family stress a selection of self- report surveys were chosen to be completed by the participant carers.

In total, 13 separate surveys are used in this study. Of these, 11 were designed to be completed by the primary caregiver, either a parent or guardian etc. of the participant. The remaining 2 questionnaires were completed by the participant i.e. the children themselves, dependant on their age. All questionnaires were filled in by the participants and family at the initial and subsequent follow up meeting, occurring approximately 3-6 months later in order to map any potential changes in carer stress over time.

A wide range of questionnaires were deemed necessary as each is focused on particular factors within the broad spectrum of family stress. As an aim of the study was to identify

specific influences of stress within a paediatric chronic illness setting, it was imperative to collect data on a wide range of potential factors so as to recognise any associations.

Although not present during the selection process of questionnaires for this study- they were identified and chosen by the M.Phil student for the academic year 2009/10- a detailed systematic review of each of the questionnaires utilised by this study has been conducted in this thesis to enable the evaluation of each of their individual/respective psychometric properties, validity and their suitability for use in this project.

Psychometrics is a field centred around the techniques and theories behind psychological measurements, for example attitudes, traits and perceived abilities. Within this area is a focus on determining the validity of these psychological measures. In this work the main type of measures used are questionnaires. These may be validated in several ways, such as content validation which ensures that each item corresponds to measuring a variable of interest. Predictive validation refers to the degree to which questionnaire responses may predict certain aspects of the subjects. Reliability is another important issue when validating a questionnaire. It refers to how accurately results would be when replicated.

4.4 Parent/Carer completed questionnaires:

4.4.1 Demographic Screen:

This standardised demographic inventory consists of 20 multiple choice items to gain an understanding of the participant family's background. Information was collected on:

- Race/ethnicity
- Family type *i.e.* both parents present, mother alone, mother with other adult, other
- Employment status
- Level of education of the mother
- Weekly family income
- Age of mother
- Number of children in household
- Other adults in the household

This screen is relevant, firstly, to allow comparison of the population characteristics between the two cohorts. This is significant in order to identify any potential confounding variables between the cohorts in the future. And secondly, it allows for the stratification of results based on different population characteristics in the later data analysis

4.4.2 Epilepsy Questionnaire:

This is a simple open-answer questionnaire which was designed by the research team. Its aim was to collect further information surrounding the potential 'seizure' events' (N.B. inverted comma's have been used to denote that, at the time of questionnaire completion, no diagnosis of these potential seizures had been made). The subject matter of the questions relates to:

- Frequency of events
- Duration of events
- Time elapsed since first seizure to presentation
- Diurnal variation
- Parents perception of these events

These are all considered potential factors which may affect the levels of stress experienced by parents. They may also serve as additional measures by which data could later be stratified.

4.4.3 Paediatric Inventory for Parents (PIP):

The PIP is a key measurement outcome as this questionnaire specifically focuses on factors affecting parental stress in relation to paediatric illness (Streisand et al., 2001). What makes it different from other tools, such as the widely used Parenting Stress Index (PSI) is it was specifically designed to be used on the parents of children with chronic conditions. Therefore it has been used to investigate parental stress in range of paediatric conditions: cancer, diabetes, sickle cell anaemia.

This questionnaire was developed in 2001 by paediatric psychologists of The Children's Hospital of Philadelphia (Streisand et al., 2003). Within the broad realm of stress it focuses specifically on the frequency of occurrence and the associated difficulty/stress caused by this. Elements of its structure were based upon Lazarus' transactional model of stress and coping (as described earlier). Although it was originally designed for usage with a paediatric oncology setting, the purposeful broadness of the questions has allowed it to be applied into a variety of other specialties.

The full version, as used in this study, comprises 42 items in total. Each is scored on two 5 point Likert scales: Frequency and difficulty. From these items four distinct subscales can be derived (Streisand et al., 2001):

1. Communications with medical/family professionals
2. Emotional functioning

3. Medical care
4. Role function

The questionnaire's key author, Streisand, conducted a large scale pilot in order to assess the validity of the PIP. Using a sample size of 126 parents within a paediatric oncology setting they investigated the psychometric properties of this instrument as a measure for stress. They found promising results regarding validity and reliability. Cronbach's α values for the frequency and difficulty domains (Streisand et al., 2001) have consistently been recorded above 0.90, indicating high levels of internal reliability. In addition, there was shown to be overall a positive correlation ($r= 0.22-0.43$) of recorded parental anxiety levels between the PIP measures and the PSI. With regard to individual subscales, there was also shown to be a higher positive correlation, with ranges of $r=0.48-0.86$ and $r=0.45-0.82$ for the frequency and difficulty scales respectively (Streisand et al., 2001). The internal consistency values reported by other studies have been collated below in Table 5.

Table 5: Previous study data investigating the internal consistency of the PIP

Study		Internal Consistency (Cronbachs α)	
Author, date	Sample size	PIP Frequency Domain	PIP Difficulty domain
(Streisand et al., 2001)	126	0.95	0.96
(Streisand et al., 2005)	134	0.94	0.95
(Ohleyer et al., 2007)	72	0.96	0.96
(Mednick et al., 2009)	20	0.93	0.94

4.4.4 Semi-Structured Stress Questionnaire (SSSQ)

The SSSQ was created by the principal study investigator, Dr. Andrew Curran, in 2009 for use in this study in order to help assess and quantify carer stress particularly with regard to paediatric epilepsy.

It comprises of 30 questions, each made up of both quantitative and qualitative elements. The questions themselves each focuses upon a different aspect of daily life. An example question taken from the SSSQ would be *'During the past 12 months how many GP/ hospital/ educational appointment appointments have you attended with your child? How stressful has this been for you?'* Some of the other areas asked about include: employment, leisure activities and household chores to name a few. The respondent first answers either 'yes' or 'no' as to whether this is stressful for them, then if they answer 'yes', they are asked to rate that stress from 0- representing very minor stress to a maximum rating of 4 signifying a great

degree of stress. Following each question is a space in which carers are invited to share any feelings they have regarding this aspect

The SSSQ design was based upon the Family Burden of Injury Interview (FBII) which is a well-validated, structured interview technique which was first used in traumatic brain injury and developed by Burgess and Taylor in 1999 (Burgess, 2008). It covers 31 items- the basis for those incorporated into the SSSQ- within an estimated timeframe of 20 minutes. Each open ended question is coded by the administrator yes/no as to whether it is stressful and then, if yes, 0-5 as to how stressful it is.

The total possible quantitative score that could be reached on this questionnaire is 96. However a cut-off score of 10 was used. Therefore carers recording <10 were deemed not stressed and those ≥ 10 stressed. This point of cut-off was decided upon by Dr. A. Curran and the first M.Phil student for this study, following personal communications with and Dr Gerry Taylor, one of the creators of the FBII.

As a result of being a novel tool, the SSSQ lacks any clinical validation and additionally there is currently no published statistical reliability information. Although it is not possible to make a direct comparison between the SSSQ and its parent questionnaire, previous validity and reliability measures for the FBII have been reasonably good as evidenced by reported alpha values of 0.90 for internal validity and Pearson product-moment correlations of $r=0.73$ for stability across 6 month follow-ups (Burgess, 2008). Of note, on a search of the literature no data was found as to inter-administrator reliability of the FBII.

4.4.5 General Health Questionnaire (GHQ-28).

The GHQ-28 is a questionnaire developed in 1978 by David Goldberg to investigate emotional stress in medical environments and as a screening tool for psychiatric morbidity (Sterling, 2011).

It consists of 28 questions which have been divided, based on factor analysis, into four distinct sub-groups: somatic symptoms, anxiety/insomnia, social dysfunction and severe depression. Each of which is answerable on a frequency scale of four: ranging from 'not at all' to 'more than usual'.

Its extreme popularity in social science and medical research has led to the emergence of different scoring methods: a basic Likert 0,1,2,3, a modified Likert 0,0,1,2, a bimodal 0,0,1,1 and chronic scoring 0,1,1,1 have all been previously used. For this study the bimodal scoring approach has been utilised instead of the original 0,1,2,3 system first suggested by Goldberg. The principal reason for this decision was due to recent work showing that the bimodal

method was shown to provide significantly more accurate findings when used for screening in non psychiatric sample populations (Friedrich et al., 2011).

Due to its widespread usage and translation into over 38 languages, a significant body of work has been generated testing the psychometric properties of this questionnaire. Research has shown strong evidence to suggest a high level reliability: test re-test reliability scores ranging from 0.78 to 0.90 and inter-rater reliability Cronbach α scores of 0.90 to 0.95 (Failde et al., 2000). Further support of this questionnaire's inclusion into this piece of research is its reported sensitivity and specificity evaluations of 79.7 and 79.2 respectively.

4.4.6 Rotter's Internal-External Locus of Control (RLoC):

This instrument was designed by Julian Rotter *et al.* in 1966 after advances in social psychology and as such, was one of the first developed 'locus of control' (as previously described in Chapter 3, section 3.1.4) (Kormanik and Rocco, 2009).

The RLoC questionnaire aims to identify whether an individual has a predominantly internal or external locus. It comprises 29 items each presented in a forced choice format in which the participant must choose one of two statements for each of the items. Of the 29 elements, only 23 are used to help determine the participant's locus. The remaining 6 are known as 'filler' items which were added by the authors in order to 'make the purpose of the test more ambiguous' (Rotter, 1966).

Scoring is binary in nature with the maximum possible score being 23. A score of 1 is given for each answer representing an external locus and therefore a higher total value indicates the extent to which the individual feels that the events around them are not determined by their own actions. Although scores towards the middle of the section indicate a healthier outlook, there are no fixed cut-off values indicating a boundary between the two groups. The reason Rotter stated for this is that locus of control is a situation specific variable. However this fact makes it more difficult for segregating the groups in a research premise. Therefore previous studies have used the median value as the cut-off point for determining internal or external LoC status.

The applications and relevance of a person's LoC is a much-debated topic in the literature. Although no firm consensus has been reached, a heavily cited seminal work by (Rotter, 1966) suggests that LoC is controlled by an equilibrium between one's 'drives for autonomy, control and social acceptance'. Therefore, it is implicit that over time LoC is subject to change: as such it has been hypothesized that situations/ life experiences may cause a shift along the internal- external scale.

Previous research using factor analysis has indicated that the RLoC demonstrates reasonable overall validity and statistically significant criterion validity in the measurement of the locus of control construct ($p < 0.05$) (Kormanik and Rocco, 2009). Several studies evaluating the internal consistency of the RLoC have reported moderate to good alpha coefficients ranging from 0.69 to 0.73. Test-retest data also shows satisfactory findings with regression coefficients between $r = 0.53$ and 0.86 .

4.4.7 Brief COPE

This is a 28 item questionnaire relating to coping strategies used by families to deal with stress. It is an abbreviated version of the original COPE questionnaire which comprises 60 items (Carver, 1997). They were both developed for use in adults with all types of disease. For this study the brief COPE was selected in order to decrease the participation burden of the study.

Each of the items aims to represent a different coping mechanism; these may be loosely be classified into functional (problem focused and emotion focused) or dysfunctional techniques (Carver, 2007). For deeper evaluation of different aspects within the realm of coping, this questionnaire can be further divided into 14 subscales, each related to factors previously shown to influence effective or ineffective coping, for example: planning, positive reframing, self-distraction etc (Carver, 1997). All are answered using a situation focused 4 point Likert scale, an example of which: 1- I haven't been doing this at all. Scores from separate subscales may be summed together in order to form aggregate scores of the main categories used in coping literature: problem focused, emotion focused and dysfunctional. This is summarised in the following table.

Table 6: Brief COPE subscale groupings (Carver, 2007)

Coping method	Subscales Used	Score range
Problem focused	Active coping	6-24

	Instrumental support Planning	
Emotion focused	Acceptance Emotional support Humour Positive reframing Religion	10-40
Dysfunctional	Behavioural disengagement Denial Self distraction Self blame Venting Substance use	12-48

Multiple studies have investigated the validity of this research tool in various different settings. Work done in 1996 by Range found good internal reliability within all subscales, with Cronbach's alpha values ranging from 0.50 to 0.90 across the 14 subscales (Range and Stringer, 1996). Test-retest investigations in recent studies have also indicated strong reliability and stability with correlation coefficients of 0.67 and 0.54 respectively (Cooper et al., 2008).

A limitation of the Brief COPE lies within the generalness of its design. While this widens its potential use into a broader range of settings and populations, it follows that it may not be specific enough to identify the necessary factors. With regard to this study it is difficult to assess the applicability of the questions to a paediatric setting. Systematic review of this questionnaires literature base revealed its use in 34 number of studies of which only 3 were within a paediatric context.

4.4.8 Family Support Scale (FSS)

This tool dates from 1986 when it was first introduced by Dunst, Trivette and Cross. It aims firstly to measure the different sources of support available to an individual and secondly to quantify how much of a support the individual perceives them to be.

The scale comprises of 18 items which each relate to a different area of potential support. The tabular format of the FSS then asks carers to rate if available, how helpful they find each of these sources of support: the possible responses range from 1-Not helpful at all to 5-Extremely helpful (Dunst and Trivette, 1986).

Scoring of this instrument is straightforward and results in a total between 0 and a maximum of 90, where higher scores indicate greater levels of support. In addition to this overall score, a series of 5 separate subscales has been proposed by the principal creators (Dunst and Trivette, 1986):

1. Formal kinship
2. Informal kinship
3. Social groups
4. Professionals
5. Professional groups

Existing research has shown the FSS to have a high level of internal consistency, reporting Cronbach's alpha values of 0.85 in an adult sample of 244 (Hanley et al., 1998) and 0.87 in a study by Sheeran (Sheeran et al., 1997) in a sample population of children with epilepsy or 'mild' cerebral palsy. Test-retest analyses were also positive with 2 large studies documenting regressions of 0.91 (Dunst and Trivette, 1986) and 0.87 (Hanley et al., 1998) over a 1 month period.

4.4.9 Family Needs Survey (FNS)

Introduced in 1988 by Bailey and Simeonsson, this instrument was designed to evaluate the perceived needs of families with children experiencing chronic disease or disability (Bailey and Simeonsson, 1988). It focuses upon functional and specific needs.

The questionnaire includes 35 items, each of which is answered on a 3 point rating system of: 1- I definitely do not need help with this, 2- not sure and 3- I definitely need help with this (Farmer et al., 2004). The questions may be separated into 6 distinct subscales based on different areas in which families identify needs. These subscales are: information needs, support needs, financial support, explaining to others, community services and family functioning.

The wide usage of the FNS has led to the variety of different scoring approaches for this instrument. The original version used a basic 1,2,3 scoring system, therefore a high score reflects a high level of unmet needs. However since then a 0,0,3 (Pit-ten Cate et al., 2002) and a 0,0,1 (Trute and Hiebert-Murphy, 2005) method have both been used. The theory behind these is that they clearly recognise only definite needs rather than assigning points for 'not sure'. In this research, following detailed conversations with chief investigator, Dr. Andrew Curran, we elected to use the bimodal 0,0,1 approach.

Evaluations of the psychometric properties of the FNS have been largely supportive. Investigations regarding internal consistency have demonstrated strong alpha coefficients of 0.91 (SEXTON et al., 1992) and 0.88 for samples of parents with children aged 0-17 years. Individual subscales produced Cronbach α values ranging from 0.65-0.86 further indicating good internal consistency (SEXTON et al., 1992). Additionally test-retest analysis showed moderate regression scores of $r=0.81$ and 0.62 for fathers and mothers respectively. Furthermore when parents recruited into Trute's study were asked their opinion of the FNS 32% rated it very helpful and 45% helpful (Trute and Hiebert-Murphy, 2005).

4.4.10 Family Adaptability and Cohesion Scale IV (FACES IV):

The FACES IV questionnaire is the fourth and most recent version of the FACES series of questionnaires which have been used in more than 1,200 published papers (Olson, 2011). They were originally designed by Burr *et al.*, the authors of the Circumplex model, to be a complementary tool to help provide a measurable outcome for family stress in a clinical context. As a result, these questionnaires are focused around the two principal orthogonal factors featured in that model (Olson et al., 1979):

1. Cohesion: This pertains to the 'emotional bonds between family members'. Its score on this questionnaire is measured on a scale ranging from disengaged to enmeshed.
2. Flexibility: This refers to the 'quality and expression of the family's leadership, organisation, roles and relationship rules' Scoring on this dimension is based on a scale ranging from rigid to chaotic.

Well-functioning families are thought to be characterised by having a balanced levels across both factors, this is determined by scoring mid-range on both the cohesion and flexibility scales (Olson, 1988) and is illustrated by Figure 12. Poor functioning families can be termed as unbalanced i.e. their scores lie on the extremes (high or low) of these scales.

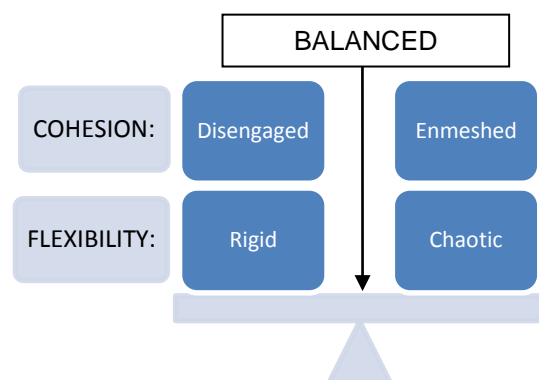


Figure 12: Illustration of the concept behind the FACES questionnaire

The FACES IV questionnaire itself comprises 42 separate statements which are each scored upon a 5 point Likert scale ranging from 1- Strongly disagree to 5- Strongly agree. These items may be subdivided into 6 family scales: 4 unbalanced and 2 balanced, which were specifically designed in order to evaluate adaptive and cohesive dimensions within families. Scoring of this tool is complex due to the amount of information the FACES IV profile aims to provide (Olson, 2010):

- Percentile scores for both the 2 balanced flexibility and cohesion scales
- Percentile scores for the 4 unbalanced scales: disengaged, enmeshed, rigid and chaotic.
- Cohesion ratio, flexibility ratio and total ratio: these seek to measure the ratio of balance to imbalance factors within the family.
- Cohesion dimension and flexibility dimension scores: to allow each family to be mapped onto the Circumplex model graph (Figure 8)
- Family communication and family satisfaction

The FACES total circumplex ratio score is of particular use in terms of this M. Phil project as a measure of one of the variables of interest: family functioning. It is calculated by dividing the average of the balanced scores by the average of the unbalanced scores. Therefore it is considered to represent how healthily the family is functioning. Interpretation of these ratio scores is shown in Table 7.

Table 7: Interpretation of FACES IV total circumplex ratio scores

Total Circumplex Ratio	Interpretation
<1	Predominantly unbalanced and unhealthy levels of functioning
1	Equal levels of healthy and unhealthy family functioning
>1	Predominantly healthy and balanced levels of family functioning

Of additional utility are the family satisfaction and family communications scales within the FACES IV. These are each comprised of 10 items scored on a 5 point Likert system (marked

from 1 through to 5) as with all the elements of this instrument. Therefore results of these scales may be easily totalled and interpreted. Scores for both scales lie between a range of 10-50 where higher scores are preferable and indicate higher levels of satisfaction or communication respectively.

Analysis regarding the tools psychometric properties is largely supportive of its inclusion into this questionnaire battery. The FACES IV confers a good level of reliability based upon documented internal consistency measures carried out in the FACES IV data manual: Cronbachs α between 0.70- 0.90 for total ratios (Olson, 2010) and α values of 0.90 and 0.94 for the family satisfaction and family communication scales respectively. This finding has been supported by other studies where reported alpha coefficients for each of the component subscales were within a range of 0.65-0.79 (Marsac and Alderfer, 2011).

Additional evidence of the validity of this instrument is the test-retest data collected from 556 participants which indicates a correlation of 0.84-0.93 over a three week period using Pearsons product-moment coefficient analysis.

Efforts have also been made to investigate the validity of this questionnaire. This has been done via convergent validity analysis. Correlations made between FACES IV and other validated family functioning instruments such as the Family Assessment Device (FAD) are detailed in Table 8.

Table 8: Convergent validity scores of FACES IV subscales and the FAD

FACES IV Subscales	Convergent Validity with the FAD	
	(Olson, 2011) n=556	(Marsac and Alderfer, 2011) n=162
Cohesion	r= -0.27- -0.82	r=0.59
Flexibility	r= 0.40- 0.85	r=0.26
Disengaged	r=0.21- 0.63	r= -0.47
Enmeshed	r= -0.20- -0.63	r= -0.09
Rigid	No data	r= -0.06
Chaotic	No data	r= -0.54

4.4.11 Strengths and Difficulties Questionnaire (SDQ)

The SDQ was developed in order to assess psychological transition in children and as a measure of emotional, behavioural and social functioning. It was originally designed by Goodman in 1997, with the extended version introduced in 1999 (Goodman, 1997). It is an extremely popular tool translated into over 60 languages and used in a wide range of chronic paediatric diseases, including: oncology, asthma and juvenile idiopathic arthritis. It been validated for completion of parents or teachers of children aged 3-16 years with an additional option of self-completion in children aged 11-16. For this research, it was always completed by the primary carer, *i.e.* parent or guardian, of the child regardless of age to allow the maximum number of comparable data-sets for this questionnaire.

The original instrument consists of 25 items, each of which is answered by one of three responses: not true, somewhat true and certainly true (Goodman, 1997). The questions correspond to different attributes of the child, including both positive and negative. Five distinct subscales may then be determined:

- | | | |
|---------------------------|---|----------|
| 1. emotional symptoms | } | Negative |
| 2. conduct problems | | |
| 3. peer problems | | |
| 4. hyperactivity symptoms | } | Positive |
| 5. pro-social behaviour | | |

For this research the extended SDQ was used, therefore in addition to the primary 25 questions a further 6 items are completed. These seek to measure the perceived impact that the child's difficulties place upon, what Goodman stated, were the key domains in the assessment of psychosocial disability namely: home life, classroom learning, friendships and leisure activities (Goodman and Scott, 1999).

Scoring of each element is dependent upon whether it is classified as a positive or negative attribute (Goodman, 1997). A different scoring system is also used by the impact section of the extended questionnaire. These methods are detailed in Tables 9 & 10 below. Final scores can then be summed for the overall total as well as the totals for each of the individual subscales. Higher scores indicate greater levels of difficulty.

Table 9: Scoring system for the original items of the SDQ

	Not true	Somewhat true	Certainly true
Negative attribute	0	1	2
Positive attribute	2	1	0

Table 10: Scoring system for the impact section of the extended SDQ

	Not at all	A little	Quite a lot	A great deal
Impact Items	0	0	1	2

This questionnaire has been shown to have reasonably strong psychometric properties. Work by Goodman *et al.* indicated higher levels of cross-informant scoring than other questionnaires and also showed it to be significantly better at predicting clinical diagnoses, such as hyperactivity disorders and inattention difficulties, than the Child Behaviour Checklist (CBCL) which is another similar instrument (Goodman and Scott, 1999).

A further, more recent study in 2001 also conducted by Goodman investigated the properties of the SDQ. By using a sample size of $n=10,438$ taken from British school children aged 5-15 nationwide implies that the results are more representative of the normal population and this study population than previous smaller scale works. It found that the overall internal consistency of the SDQ was good, with Cronbach's α reported as 0.80 (Goodman, 2001).

Several investigations have also shown high correlations in test-retest analysis. A Swedish pilot study reported a correlation of 0.96 over a 2 week period (Smedje *et al.*, 1999) whilst a British sample documented intra class correlations of 0.85 over a duration of 3 weeks (Beretvas *et al.*, 2008) (Goodman and Scott, 1999).

4.5 Child completed questionnaires:

4.5.1 Self Image Profile for Children/Adolescents (SIP-C and SIP-A).

This questionnaire was first introduced by Butler in 2001. It was developed to be self-completed by the participant child/ young adult in order to help quantify their perceived level of self-image and self esteem. These are considered important variables as a child or adolescents construct of self has be shown to be fundamental to their psychological wellbeing (Thomas *et al.*, 2011).

There are currently two versions available to allow for differing language and comprehension abilities between children of different ages: SIP-C for children 7-11 years of age and SIP-A ages 12 to 16 (Children aged 6 or below who were included in this study were not required to complete this tool).

Both forms of this survey comprise 25 items corresponding to a personal attribute or characteristic (Butler and Gasson, 2005). Of these, 12 are classified as positive, 12 as negative with 1 neutral item. To complete the tool participants are asked to make 2 ratings for each of the items (Butler and Gasson, 2005). Firstly, 'how I consider myself to be' and secondly 'how I would like to be.' These ratings are both scored upon a Likert scale which ranges from: 0- not at all to 6- very much.

This questionnaire is interpreted by a positive self image score (ranging from 0-72) comprised from the 12 positive items, a negative self image score (again spanning 0-72) based upon summation of the 12 negative items and a 'sense of difference score' based on the single neutral item which may have a total of 6 and a self esteem measure which compares the difference between the 2 rounds of scorings (Butler, 2001). A higher positive score reflects that the child views them self as having mostly positive attributes whilst a higher negative score suggests that the individual sees them self as having a more negative attributes (Butler, 2001). The size of the sense of difference score is thought to reflect the degree of which the child or adolescent views them self as being different. Higher scores imply that the individual in question feels separate from their peers (Butler, 2001).

Previous literature on the psychometric properties of this instrument indicate that the SIP-C and SIP-A both demonstrate fair to moderate degrees of internal consistency. Overall alpha coefficients for both the child and adolescent versions were recorded as 0.69 and 0.79 respectively (Butler and Gasson, 2005). In addition, when analysing the positive and negative elements separately work by Butler and Gasson, based on samples of children from primary and secondary schools in Leeds showed Cronbachs alpha values for SIP-C positive and negative $\alpha=0.69$ and for SIP-A positive items 0.69 and negative items 0.79 (Butler and Gasson, 2005).

4.5.2 Paediatric Quality of Life Inventory (PedsQL 4.0):

This questionnaire, currently on its fourth version, was designed by Varni, Seid and Kurtin to investigate a child's health related quality of life (HRQOL) (Varni et al., 2005). This tool may be self-completed by children aged 6-18 years to help assess their perception of their own quality of life. Despite its origins in paediatric oncology settings, the PedsQL is a generic disease based tool which incorporates both disease-specific and generic health elements. It has been proposed that this quality makes it particularly suitable for investigating HRQOL in populations where a diagnosis has not yet been made.

In total this questionnaire consists of 23 items, each in the form of statements. Children are then asked to score them all based upon how often they think/feel that way using a 5 point

Likert scale anchored by: 0- never to 4 all the time. From the total 23, certain elements correspond to different areas encompassed within HRQOL, these can then be used to form 4 distinct subscales(Varni et al., 2005):

1. Physical functioning
2. Emotional functioning
3. Social functioning
4. School functioning

Scoring of this questionnaire is straight forward, summing the Likert numerical values for each response. These provide an overall score out of a maximum 92. In addition to this total, individual subscale scores may be calculated based upon their corresponding items.

Previous studies have recommended the PedsQL for measuring health-related quality of life purposes. Additional research has validated this instrument for use in children aged ≥ 6 years and shown this tool to maintain a high level of internal reliability: Cronbach's α values were 0.88 and 0.90 for self and proxy-reporting respectively. Additionally, summary self scores all exceeded 0.70 for each subscale (Varni and Limbers, 2009, Davis et al., 2010).

4.6 Discussion and Personal Critique of Questionnaires

Overall, I have complete confidence in the selected questionnaires, based on the existing literature and personal experience of using them. However, from this experience there are several potential improvements that I feel could be implemented in the future.

One example of this lies with the Rotter's LoC measure. Although it still has its place, the fields of social and clinical psychology have greatly evolved since the development of Rotter's LoC and some of the questions are slightly dated and may not be considered strictly relevant. In particular, items 3 and 22 (which focus on politics and war), were frequently commented on to me, by the parents completing the questionnaire as to why they were being asked their opinions on such topics. In the 40 plus years since the creation of Rotter's LoC, several other measures have been introduced to measure locus. Of note, are health specific locus of control questionnaires (Luszczynska and Schwarzer, 2005). An example of this is the 'Health Locus of Control (HLC) scale, which was based upon the original Rotter's tool. This questionnaire is shorter using only 11 items and has show good internal consistency with an alpha coefficient of 0.72 and a correlation of $r=0.33$ with Rotter's LoC (Wallston et al., 1976).

Were this study being designed now, I would consider using this tool because as noted in the literature there are difficulties associated with using a general measure to determine health specific locus of control and the sensitivity of the questionnaire may be diminished (Wallston et al., 1976).

Upon first inspection, it was not immediately clear as to why certain questionnaires were included in this study. The FNS for example, although shown to be useful and valid questionnaires does not automatically fit into the family stress nature of the study. However, on a practical point it helps gain an insight into areas in which families may find the systems and resources at AHCH could be improved.

Whilst reviewing the existing literature on stress and paediatric epilepsy, it became apparent that some of the questionnaires being used in other research were comparable to those used in this piece of research in terms of validity and reliability scores. Following this, further investigation was done into these specific tools with regard to their potential suitability to this project and why some were chosen over others. Over the course of the thesis, the merits and weaknesses of each will be further discussed with future possibilities and adaptations discussed predominantly in Chapter 5.

The PSI is a screening and diagnostic tool that was designed to measure stress levels within parent-child systems (Loyd and Abidin, 1985). First developed by Abidin, it is now one of the most long-standing and widely used stress questionnaires. This questionnaire may be used either in its original format 120 items or in its short form of 36 questions (which would be most appropriate in a study such as this). A total score of the reduced format may be calculated based on summing all responses but it may also be segregated into 3 key subscales, namely: parental distress, parent-child dysfunctional interaction and difficult child (Reitman et al., 2002).

It has repeatedly been demonstrated to have high levels of internal consistency, as evidenced by Cronbach alpha values of 0.95 for the total score and coefficients ranging from 0.88 to 0.95 for each of its subscales (Reitman et al., 2002). Factor analysis has also revealed encouraging results with a comparative fit index of 0.90 (Haskett et al., 2006). Additionally strong correlations have been documented between the short form PSI and other validated measures such as the Symptom Check List (Haskett et al., 2006).

Upon discussing the strengths and potential of the PSI with supervisor and lead investigator Dr A Curran, it was revealed that the PSI was in fact the first choice for use in this project. However, on presentation to the Alder Hey steering committee, an objection to its use was raised by one of the psychologists on the board who felt it to be too distressing for families (based upon one occasion where she witnessed a mother cry whilst completing it.)

Chapter 5: The Epilepsy Stress Study

The following sections of this thesis are focused on presenting the Epilepsy Stress Study as carried out by this author (and previous colleagues) and the results obtained through this work.

5.1 Study Title

Correlates of stress in carers of children newly diagnosed with epilepsy

5.2 Aims

To identify some of the key variables that affect how a family deals with the stress of their child's new diagnosis of epilepsy. To delineate how that stress changes over time and in relation to disease or carer variables. To better understand potential factors causing increased stress with a view to being able to help alleviate them.

The prime objective of this study is to have a greater understanding of the families' experience of the time interval around initial diagnosis and any particular factors associated with greater levels of stress. With this enhanced knowledge it would then be possible to audit existing programmes available to provide improved parental and family support. Following on from that, guide the development of new resource measures to provide a better service to these families to help secure not only a better outcome for the individual child but also an improved wellbeing of the family group as a whole.

A future aim, on the completion of this work, is its publication in a scientific paediatric journal which would allow for the widespread distribution of any significant results/trends identified. In turn, this will allow a broad range of medical professionals working within paediatrics to have increased knowledge of this subject area when making the best evidence-based decisions for this cohort of patients and families.

5.3 Hypotheses

Three broad null hypotheses were drawn up at the beginning of this academic year as an overreaching focus for the research. These were:

1. There is no difference in carer stress scores between cohorts at T0*
2. There is no difference in carer stress scores between cohorts at T6*
3. There is no association between carer stress and the other variables

*Definitions of the cohorts and their allocations are detailed in Section 5.5.2

5.4 Outcome Variables

During the initial phases in the design of the study, a set of variables were identified as being of interest to the investigation of stress correlates in paediatric epilepsy, these are shown in Table 11 below.

Table 11: Summary of the variables under investigation

Carer Variables:	Child Variables:
Stress	'Seizure' Event History
Coping	Quality of Life
Supports	Self esteem
Needs	Self Image
Health	Behaviour
Locus of Control	
Family Functioning	
Family Communication	

5.5 Methodology

5.5.1 Systematic Review of the Literature

As a result of taking over the study in its third year, an extensive search of the existing knowledge base surrounding the progression of family stress theory was carried out. This was done by using medical and psychology based search engines such as: PubMed, PsychInfo, Scopus and Ovid. Initially broad search terms were used and results limited to review articles so as to start with a good general background of the topic. Further refinements using the MeSH headings to expand terms, allowed the papers that may have initially been missed to be retrieved. Following this, more specific criteria were employed in order to identify the most current and relevant literature for the study.

In addition to researching family stress, a detailed systematic review of each of the questionnaire tools included in the study was undertaken.

To analyse the literature in a logical and structured manner the CASP approach was employed. This tool provides an ordered framework to assist in the evaluation of scientific

papers by systematically breaking them down to focus on the key questions that need to be answered in order to gain an insight into the relative strength and validity of the conclusions made (Health), 2012).

5.5.2 Study design:

This is a longitudinal controlled cohort study. It is multiple questionnaire based in nature, where, as previously stated, 9 surveys are completed by the primary carer and 2 others are completed by the affected child. Questionnaire data from each participating family is collected at two intervals: At time of recruitment i.e. at 0 months and at the child's follow up clinic appointment usually occurring approximately 6 months later.

The study group was divided retrospectively into the following two cohorts:

- Group 1: Children who following their first presentation are diagnosed with epilepsy. Diagnosis will be based on satisfying the epidemiological definition of epilepsy outlined by the International League against Epilepsy as 'a condition characterized by recurrent (two or more) epileptic seizures, unprovoked by any immediate identified cause (Berg et al., 2010).
- Group 2: Children who following their initial presentation are found not to have epilepsy. They may either be discharged from clinic as healthy or be referred to another paediatric specialty for a non-neurological diagnosis of the events.

The two cohorts will be directly comparable as each will have had similar experiences up until the point of diagnosis, therefore the children without epilepsy can act as the control group for the children with epilepsy. As neither the families themselves nor the recruiting researcher know the diagnosis of epilepsy at the time of recruitment there would be single blinding which should minimise any event of observer bias. Standardisation will also be carried out as per participant demography to allow for any potential confounding variables.

The study uses the popular multiple questionnaire based design. An example of which, as specifically mentioned in the original study proposal, is the work done by Taylor *et al.* in their research regarding traumatic brain injury (TBI) in children. This work investigated bidirectional child-family relationships as affected following TBI, collecting data at 0, 6 and 12 month intervals. By using multiple questionnaires completed by child and parent, later statistical analysis using Structural equation modelling (SEM) and path analysis revealed statistically significant results. By using a similar structure in the design of this study it is hoped that equally significant and reliable results may be found.

5.5.3 Justification of Study Design

Modern survey-based designs date back to Victorian Britain (Bowling, 2009). It is an effective and popular method of collecting descriptive and analytical information from a sample population of interest. As stated, this study is also longitudinal in nature which also allows for the investigation of any potential causal relationships between variables (Bowling, 2009).

The use of a questionnaire battery may be justified by the popularity of this method in the recent literature and the quality of information that it yields. Previous research into paediatric epilepsy, such as the SANAD randomised control trial also included a questionnaire battery as part of their data collection method for quality of life measures (Marson et al., 2007).

From extensive searches of the literature, as detailed in chapter 4, previous research methods into parent/carer stress in childhood illness has followed a variety of methods mainly quantitative or mixed data collection. Although a solely qualitative approach could have been undertaken, for example via either cohort interviews or focus groups, it would not be possible to gain an objective measure of the carer's stress or of the other covariates. This study design uses primarily quantitative questionnaires as well as some qualitative elements

Random sampling was not possible for this study due to the recruitment setting and the already limited number of possible participants. Observer bias based upon cohort status was however limited based upon the retrospective nature of determining the cohort status of each individual.

5.5.4 Questions of Interest

In addition to the broad hypotheses as described above, the following more specific research aims were developed during this final year of the study by this author. These were decided upon prior to the completion of data collection and statistical analysis following discussions with the study's statistical advisor, in order to avoid the suggestion of data dredging (Kent, 2001).

Data dredging refers to the false identification of a correlation or significant finding as a result of trawling a large database, such as that of produced by this study. It is therefore hoped that this will have helped eliminate a potential source of bias.

My specific research aims were to:

- Compare of the (average) scores for each of the stress questionnaires at baseline: over total population and between the two cohorts

- Determine any correlation between each of the individual stress questionnaires i.e. consistently high/low across all surveys.
- Compare scores for each of the questionnaires at follow up overall and between the two cohorts.
- Compare changes in stress scores over time i.e. between T:0 and T:1 (average time between recruitment and follow up)
- Determine any relationship to number of seizures and correlations to stress scores across total population and between cohorts.
- Assess any correlation between carer stress scores when compared to:
 - The seizure frequency of the participant child,
 - The age of the participant child,
 - The number of children in the family
 - The carers age.
 - The self-perceived family ethnicity
- Determine whether carer stress scores correlate to their child's reported HRQOL scores.
- Compare stress scores (total population and then between cohorts) to:
 - Rotters Locus of Control: Are there any significant differences in stress scores between families with internal or external loci of control
 - FSS: Does the amount of social support as quantified by the FSS questionnaire have any influence upon stress scores.
 - FACES IV: Is there a significant difference in the stress scores of families identified as being either balanced or unbalanced.
 - COPE: Do those who self report coping mechanisms have different levels of stress compared to those that do not.
 - GHQ: Does the carer's self perceived health, identified by the GHQ-28, have any correlation with their reported stress scores.

5.5.5 Outcome Measures

The outcome measures for this research are the thirteen self-complete questionnaires described in Chapter 4 of this thesis. Each of these questionnaires focuses upon and investigates a different potential covariate that may be associated with the multi-factorial stress concept. A brief summary of each of these is shown in Table 12.

Table 12: Summary of study variables and their respective measures

	Study Variable	Questionnaire	Summary of Key Concept	Reliability and Validity	Method of Scoring
Carer Variable	Stress levels	1 PIP 2 SSSQ	2 domains: the frequency of a particular aspect of health/ behaviour and the associated difficulty of it. A structured interview investigating different potential contributing factors to stress.	Cronbachs α 0.96 and 0.95. R= 0.48-0.86 between domains Not available (FBII α value 0.90)	Total score possible 150, for each domain. >90 indicates increased level of stress Scored out of a maximum of 96. Cut-off score of 10. Greater than 10 indicative of Stress
	Health	GHQ-28	Collects information on carer mental health focusing on 4 key areas: somatic symptoms, anxiety/insomnia, social dysfunction and depression symptoms.	Cronbachs α 0.90- 0.95. Test-retest validity 0.78- 0.90.	4 point likert scale each item scored as: 0,0,1,1 Scores > 5, out of a total of 16 are used as a cut-off.
	Locus of Control	RLoC	Investigating personality aspects and the level to which a person/family believes that the situation around them is affected by their own actions	Reported α coefficients of 0.69-0.73 Test-retest correlations of 0.53-0.86.	Categorical. Scores are out of 23. The median value of the samples are used for determining the cut-off between internal and external loci.
	Support	FSS	Measures the total level of support received by a person and where they come from. Divided into: formal, informal, professional and professional groups support	Cronbachs α 0.85 for total FSS score and 0.48-0.87 for subscales. Test-retest correlations of 0.73.	Maximum possible score of 90. No categorical cut-off values. Higher scores indicate greater support.
	Needs	FNS	Definite needs are scored by parents in 3 key categories:	Internal consistency as per α values of:	Using the modified bimodal scoring

			information needs, resource needs and counselling needs	0.88-0.91. 2 week test-retest correlation of 0.79-0.90	method maximum possible score is 35. Higher scores imply greater levels of needs
	Family Functioning	FACES IV	Identifying the levels of flexibility and cohesion within families.	Subscale internal consistencies of $\alpha=0.77-0.94$. Test-retest correlations between 0.83-0.93	62 items each scored from 1-5. Using the FACES excel programme individual scale ratios and overall ratios are calculated to be used in comparisons
	Family Communication	FACES IV	A 10 item scale rating family communication.	As above	Raw scores are converted into total percentages.
Child Variable	'Fit' History	'Fit' Questionnaire	Collecting data on seizure characteristics	Not applicable	Majority of items are qualitative. Seizure history scored as a continuous variable
	Quality of Life	PedsQL	Child completed quality of life measure. Gives a total score but also creates 4 subscales: physical function, emotional function, social function and school function.	α values of 0.86 for total scores and ranging from 0.68-0.88 for each subscale.	Total scores are from a range 0-100 with higher scores indicating a greater HRQOL.
	Self Image Self Esteem	SIP-A/C	Measures child self image based on the positive & negative attributes that the child perceives that they have	Cronbachs α of 0.69-0.79	Positive and negative SI scales range from 0-72. Sense of difference measures from 0-6.
	Behaviour	SDQ	A proxy measure of behaviour filled by the child's parent. Seeks to quantify	α coefficient of 0.73 Test-retest correlations of	Maximum score of 40. Scores of 14-16 deemed borderline

			the difficult behaviours of a child	0.62	behaviour and those >16 abnormal.
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5.5.6 Candidate selection:

All referrals to the paediatric neurology team at Alder Hey Children's Hospital, including internal referrals as well as those from primary care/ general paediatrics, were examined as potential study participants.

Inclusion Criteria:

1. The child's age is between 0 and 16 years.
2. The child is a registered patient of Alder Hey Children's Hospital
3. The parent/carer is deemed competent, both to understand the nature of the study and what is required of their family should they consent to participate.
4. Referral to an Alder Hey outpatient clinic (either: Neurology, General Paediatrics or First Fit clinics) due to a suspected seizure disorder, of which epilepsy could be a potential cause.

Exclusion Criteria:

1. The child has prior diagnoses/investigations for epilepsy or is currently taking regular AEDs.
2. The suspected seizure disorder was precipitated by an discrete event or trauma.
3. The child has another medical diagnosis, either physiological or psychological which is severe or relapsing-remitting in nature.
4. The carer/family do not speak fluent English.

5.5.7 Justification for the Candidate Selection Criteria

Inclusion:

1. An age range of 0-16 years is valid for this study as the participants would still be seen as a child, from medical and legal perspectives and therefore fit the age specifications to be diagnosed with paediatric epilepsy, which is the particular interest of this work.

2. The child must be a registered patient of AHCH due to the ethics proposal of this research, in which it was clearly stated as a single-centre study. This criterion is also justified on a practical/funding-based level. Only one person (a M.Phil student) was ever in charge of recruitment at time. Clinics, from which it was possible to recruit participant from, at Alder Hey occur 5 days a week. Several of these overlap each other. Therefore, in some cases it was already difficult to reach all of the potential recruits at only one centre, let alone another hospital.
3. Understanding and competence of the child's carer is a key part of obtaining valid consent, therefore it is a necessity for the selection process of this study. Both from an ethical stand point but also in maintaining the best interests of the child and family.
4. Attendance at an outpatient clinic is a necessary criterion as this is where recruitment occurred.

Exclusion:

1. A prior diagnosis of epilepsy is an essential distinction as the express concern of this study is the impact of a new diagnosis of epilepsy on the family. It is reasonable to assume that a family who has already had exposure to epilepsy and has already had time to come to terms with the diagnosis may not experience the same levels of stress as those for whom this is a new experience. Therefore in order to reduce any potential skew/bias in the results, these people were excluded from recruitment.
2. Epilepsy as a secondary occurrence following an event or trauma e.g. traumatic brain injury or ischaemia could potentially impact the results. This is because these events are likely to have their own associated stress, leading to a potential bias of the results.
3. The presence of a concurrent relapsing-remitting medical condition is a relevant exclusion criterion as it is similar to the disease course of epilepsy. Therefore, on interpretation of the questionnaire results it would be difficult to determine whether the levels of stress and associated covariates would be due to the epilepsy. The initial proposal for this study suggested excluding children if they had an existing medical condition. However, children with epilepsy are, by nature, more likely to have a comorbid medical condition and therefore, it was felt that such a criterion would not only limit but also bias the selection process of the study. The presence of a stable chronic condition was not felt to impact the scoring of the questionnaires as its constant nature would imply that its associated stress levels would be the same between the two data collection points.
4. Although some of the questionnaires have been validated for their translations into other languages this is not the case for all. On this basis it was decided not to recruit carers who do not speak English. This was also due in part to the non-funded nature

of the study and the expenses involved in buying the questionnaires in different languages. It will however be acknowledged in the discussion

5.5.8 Selection Process

Upon satisfying the aforementioned criteria, these identified persons were sent participant invitation packs in the post. These packs comprised:

- A participation invitation letter,
- A parent information pack,
- A children's information pack. Two child information packs were developed, one for children less than 10 years of age and one for those greater than 10 years. Content included in each was similar although presented in a more appropriate way for the different age ranges. Children less than 6 years were not sent separate information packs.

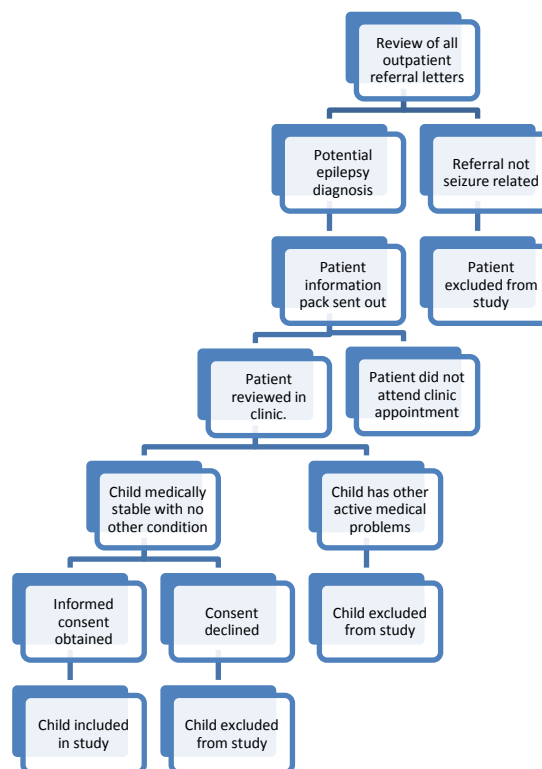


Figure 13: Flow Chart representing Recruitment Process

First meeting:

The initial meeting coincided with the child's first clinic appointment at the neurology department. Following their appointment the researcher met with the child and their parents to discuss the study. Once informed consent was obtained, a family member (primary caregiver)

was asked to complete the required surveys. The researcher remained with the parents at this time in order to answer any queries the parents had and ensure all parts of the surveys were completed. Depending on the age of the child they completed the appropriate questionnaires for themselves. Table 13 indicates the questionnaires completed by each family member.

Table 13: A Summary of the Questionnaire Battery Completed by the Different Study Participants

Participant:		Questionnaires Completed:
Carer		Paediatric Inventory for Parents Semi-Structured Stress Questionnaire FACES IV Strengths and Difficulties Family Support Scale Family Needs Survey Rotter's Locus of Control General Health Questionnaire 28 Brief COPE Inventory
Child:	0-5 years	None
	6 years	Paediatric Quality of Life Inventory
	7-11 years	Paediatric Quality of Life Inventory Self-Image Profile for Children
	12-16 years	Paediatric Quality of Life Inventory Self-Image Profile for Adolescents

Follow up meeting:

After the first meeting participants were followed up between 3 to 6 months later. The majority of patients were followed up at the neurology outpatient department at the same time as their return clinic appointments. At this time parents (preferably the same individual who completed the surveys at the initial meeting) completed the same surveys for a second time. If appropriate, the child also completed the same questionnaires for a second time.

In some cases, currently 9, it was not possible to have a follow up meeting with the participant family. Reason for this include: child requires no further outpatient appointments at Alder Hey, the family did not attend their appointment, researcher unavailability. When this occurred, a questionnaire pack was sent to the family's recorded address with a set of instructions for filling them in as well as a pre-addressed envelope to return the completed questionnaires.

5.5.9 Data Management:

Collected questionnaire data was transferred onto excel spreadsheets in order to collate all of the information. It was also copied onto an SPSS database to allow more complex statistical analyses to be carried out.

The majority of the data is quantitative as the scoring techniques employed by the chosen questionnaires were predominantly Likert-based scales. Some of the surveys however did include qualitative answer fields. Due to the aforementioned present lack of funding for qualitative analysis the paper copies of all the original completed questionnaires have been safely stored in locked cabinets for use in this purpose at a later date.

5.5.10 Ethics:

Research Ethics Committee

The study protocol was approved by the North West 3 Research Ethics Committee- Liverpool East in January 2010. REC reference number: 09/H1002/91. Following this, amendments were made to the original protocol and were subsequently submitted and accepted by the Research Ethics Committee on 2 separate occasions, as illustrated by Table 14.

Table 14: Compliance and Correspondance with the Research Ethics Committee

Document	Version	Date of Approval
Application	1.0	20/01/2010
Protocol		20/01/2010
	Ammendments (Minor)	18/05/2010
	Ammendments (Substantial)	16/09/2010
Participant Invitation Letter	1.2	09/02/2010
Participant Information Packs	1.2	09/02/2010

The first amendment to be made was an adjustment to the inclusion criteria of the study. Initially it was stated that only children between the ages of 6-16 years of age would be eligible to take part. However, during the first few months of the study it was clear that recruitment targets were not being met. Therefore, it was put forward that the age range be altered to 0-16 years. And, in the situation of a child being recruited being aged 0-6 years, the 2 questionnaires to be completed by the child would not be given.

Consent

Before inclusion into the study, informed consent was required from all potential participants. Study information booklets were posted to potential participants in advance of their clinic appointment, allowing plenty of time to read and process all the information. These information packs also contained contact phone numbers for both the M. Phil student at the time as well as Dr Andrew Curran (consultant paediatric neurologist and lead investigator for this study) should either the parents or child have any questions pertaining to the study. These measures help satisfy GMC requirements for consent in research (Council, 2010).

Following their clinic appointment, participants were approached in person where a study member was present to answer any questions. If the participants family expressed interest in taking part, consent forms were signed in duplicate (One for the study records and one for the families to keep). In addition, children greater than 6 years of age were given separate age appropriate child consent forms: one for ages 6-10 years and one for >10 years. These children were also able to ask any questions or refuse before consenting to participate in the study.

Confidentiality

Confidentiality was an important consideration for this study (Council, 2010). Patient records (paper and computerised) were consulted only when necessary. Completed forms and questionnaires were kept in locked cabinets and all computerised patient data was encrypted as per Alder Hey computer services department requirements. Furthermore, for patient analysis all individual identifiable information was made anonymous.

5.5.11 Statistical analysis:

Primary statistical input during the proposal development was carried out in conjunction with Dr. Steven Lane, a bio-statistician from the University of Liverpool. This was done to ensure the most appropriate and effective statistical methods to achieve the study aims were chosen and that the data collected would allow this.

Unfortunately, during the planning stages of this study a power calculation or equivalent was not undertaken. It was recognised and explained at the time in the protocol that it was due to the 'relatively novel' design of the study i.e. it was not then possible to find suitable comparative data/figures with which to conduct an adequate alpha value calculation. It is important however to notice that this may have had a subsequent effect when it came to recruitment goals and drawing statistically significant conclusions from the data.

It was originally decided for the statistical analysis to be centred on Structural Equation Modelling (SEM). This is a popular technique used for estimating causal relations based on statistical and qualitative assumptions. It combines aspects from factor analysis, path analysis and regression analysis. This particular method was chosen for the study, due to its strength in testing overall models compared to multiple regression analysis with tests only independent coefficients.

Due to study circumstances, the use of SEM for data analysis had to be abandoned. This was due to insufficient sample size which would prevent the likelihood of obtaining any statistically significant results. Therefore alternative statistical methods were decided upon to conduct multivariate analysis.

5.6 Results

The subsequent section of this thesis presents the data collected by this study. Firstly the results and analysis of each individual questionnaire are reported with the associated comparative analyses conducted between the cohorts. Following this, inter-variable correlation analyses are also presented.

5.6.1 Recruitment Data

Overall, 196 patient information packs were sent out to families. From this a total of 88 families were recruited into the study. This gives a recruitment rate of 45%. Reasons for some families not being recruited into the study and the frequency of this occurrence have been tabulated below.

Table 15: Summary of the Reasons why Families Were Not Recruited Into the Study

Reason for Non-Recruitment	Frequency (Percentage to 2 sig.fig.)
Family declined consent	27 (25%)
Family DNA clinic appointment	39 (36%)
Study investigator unavailability	17 (16%)
Other	25 (23%)

Follow-up appointments are still ongoing, but at the time of writing this thesis, 43 of the participant families had completed the second battery of questionnaires. The range of time between recruitment and follow-up was 5.2 to 8.9 months with a mean of 6.4 months. However, due to incomplete data fields, 4 carers were excluded. This results in 39 paired datasets.

Table 16: Breakdown of Carers Completing Each Timepoint.

Appointment	Frequency
1 st Appointment only	45
• Proportion lost to follow up	17
• Proportion ongoing i.e. not reached 6 month interval yet.	28
1 st and 2 nd Appointments completed	43 (N.B. incomplete data fields meant 4 datasets were excluded)

5.6.2 Study Population Demographics

Demographic data from all participating families has been compiled into Table 17 below. The age of the children in the study ranged from 0-15 years. Child gender ratios were comparable both overall and between cohorts

With regard to the caregivers' variables, 82% of those completing the questionnaire were the mothers compared to 15% of fathers. The age range of the primary carers ranged from 24-49 years, with a mean age of 34.9. The majority of caregivers reported their ethnicity as white/Caucasian with only 2 identifying with other ethnic groups. The level of education attained by the carers was diverse; however a reported 85% completed secondary school. 74% of the primary caregivers were employed at time of completing the questionnaires.

The structure of the participating families was completely nuclear in nature. 85% of families had 2 parents present, although this includes step parents and adoptive parents in addition to biological parents. Number of children in the family ranged from 1-4 with the mode for the total sample and for both the cohorts being 2 children.

Table 17: Demographic data of the study population

Variable	Total Sample (n=39)	Epilepsy (n=18)	Non Epilepsy (n=21)	Group Differences (Chi square/ Fishers Exact test)
Age of Child	7.6	7.4	7.7	0.957
Gender of Child (M/F)	20/19	9/9	12/9	0.666
Relationship of primary carer to child:				
Mother	32	15	16	0.591
Father	6	3	4	
Other	1	0	1	
Age of primary carer	34.9	34.6	35.2	0.477
Ethnicity:				0.880
White/Caucasian	37	17	20	
Asian	1	0	1	
Black/Afro Caribbean	1	1	0	
Education level of carer:				
Some school	6	3	3	0.064
Finished school	9	2	7	
Some college	6	6	0	
Finished college	11	4	7	
Higher degree	7	3	4	
Employment status of carer:				
Employed	29	15	14	0.235
Unemployed	10	3	7	
Family Structure:				
2 parents (bio)	25	11	14	0.614
2 parents (step)	4	1	3	
2 parents (adoptive)	4	2	2	
Single parent	6	4	2	
Number of children in the household:				
1	6	1	5	0.271
2	21	11	10	
3	8	5	3	
4	4	1	3	

Based upon the table above, it is apparent that none of the statistical tests reported values <0.05. This suggests that there are no significant differences between the population demographics of the 2 cohorts. Therefore it is justified to conduct comparative analyses between the cohorts for the measured covariates.

5.6.3 'Seizure' Characteristics Data

The data below, Table 18, was that collected from the Seizure event Questionnaire. Results for only 3 out of the total 7 items are detailed below. This is because, as previously stated, qualitative data analysis will not be undertaken for this thesis. Funding for this aspect of the results is still being sought.

Table 18: Data from 'Fit' characteristics questionnaire

Variable	Total Sample (n=39)	Epilepsy (n=18)	Non-Epilepsy (n=21)	Group Differences (Chi square/ Fishers Exact test)
Fit frequency:				
1-4 x total	16	6	10	} 0.096
1-2 x month	16	10	8	
1-2 x week	2	0	2	
3-5 x week	2	1	1	
Everyday	1	1	0	
Period of time since onset:				
1-2 months	12	9	3	} 0.085
3-4 months	16	5	11	
5-6 months	3	2	1	
>6 months	8	2	6	
Previous investigations before 1st clinic appointment	3	2	1	0.382

Table 19: Further characteristics between the 2 cohorts and the timing of their study appointments

	Epilepsy Cohort	Non Epilepsy Cohort
Mean age	7.4	7.7
Median age	7	7
Range of ages	4-16	1-15
Diagnosis	13 generalised epilepsies 5 Focal epilepsies	7 vasovagal syncope 4 migraine 1 tics and mannerisms 9 unknown. But confirmed not epilepsy.
Appointment timing % <ul style="list-style-type: none"> • Prior to review by the clinician • Post review by the clinician • Unknown 	51% 23% 26%	54% 18% 28%

Table 19 above provides further detail to the characteristics of the study participants. The mean and median ages between the 2 cohorts were similar with no significant difference. Additionally, the range of ages was distinctly overlapped although the lowest age in the epilepsy cohort was 4 compared to 1 in the non epilepsy group. Again this was not found to be statistically different. Because these groups were not following a normal distribution standard deviation calculations were deemed to be inappropriate by the statistical advisor.

The diagnoses received by each child were also collected. The epilepsy contingent was only divided into generalised and focal categories based on the original members who produced the study design. Unfortunately subgroups of epilepsy type were not recorded at the time and ethics protocol inhibited further checking.

As tabulated above, the majority of participant appointments were achieved before meeting with the relevant clinician. Differences between timing of appointment were also statistically analysed and found not to be significant. However, for approximately a quarter of each cohort the timing of their appointments remains unknown. These issues are mentioned in further detail in the discussion section of this work.

5.6.4 Paediatric Inventory for Parents Analysis

Total frequency and difficulty domain scores

The histograms below indicate the spread of data across the 2 domains of the PIP at T0. The orange reference line on the x-axes represent a score of 90, which was used in concordance with the existing literature as the cut-off score for indicating whether a parent/carer is experiencing greater than normal levels of stress.

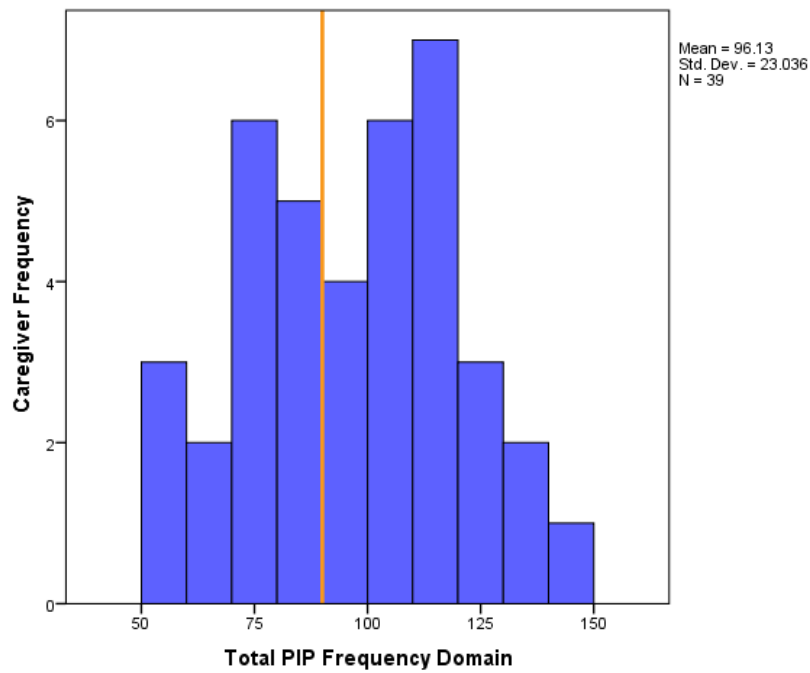


Figure 14: Histogram of total caregivers PIP-f scores at T0

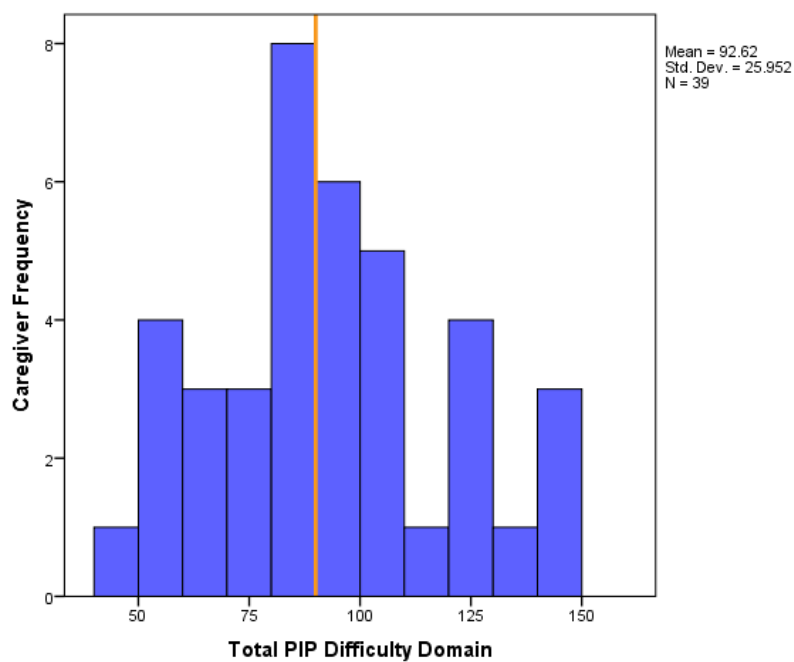


Figure 15: Histogram of total caregivers PIP-d scores at T0

As seen in Figures 14 and 15 there is a wide spread of scores on both of the stress domains included in the PIP at T0. They show that at initial presentation, 59% of carers are scoring above normal levels of stress on the PIP frequency domain and 56% on the difficulty domain.

Correlation between Frequency and Difficulty Domains

Correlation analyses were then carried out, to assess the association between the difficulty and frequency domain scores of the PIP at both time points. This was to determine whether the carers were scoring highly on both domains.

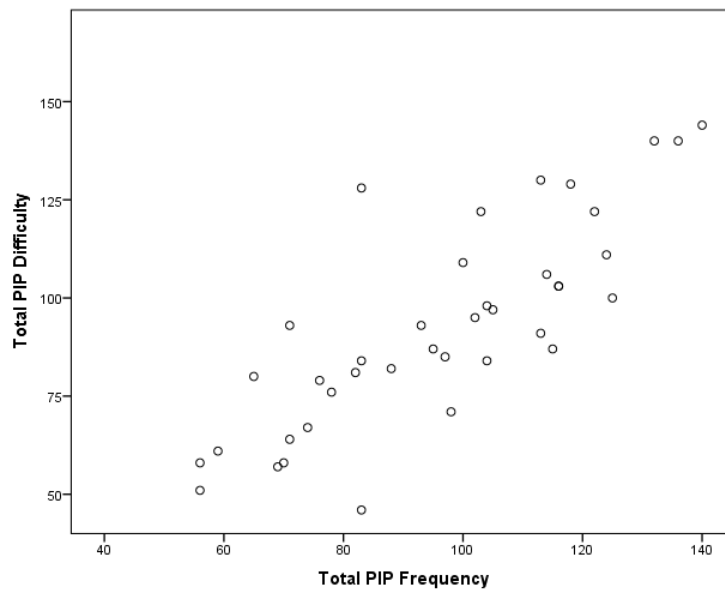


Figure 16: Correlation of PIP-f and PIP-d domain scores of total sample population at T0

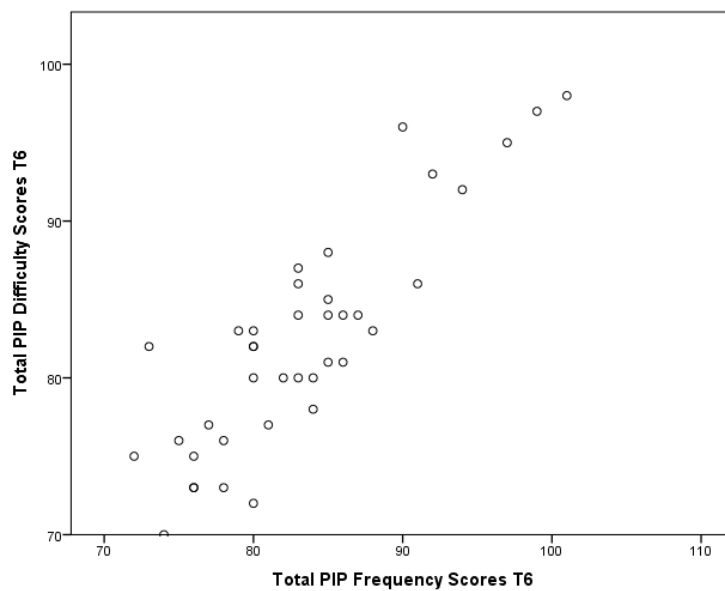


Figure 17: Correlation of PIP-f and PIP-d domain scores for total sample population at T6

The preceding scatter graphs demonstrate that there was indeed an association between carers scoring of the 2 PIP domains. Spearman's rank tests confirmed this observation by calculating correlation coefficients of 0.65 and 0.75 for the T0 and T6 data respectively. This indicates that at both time points there was a strong positive correlation between carers stress associated with the frequency and difficulty of the suspected seizure events.

Cohort Analyses

In addition to analyses of the overall sample population, comparative analyses of the total PIP frequency and difficulty domain scores were carried out by epilepsy status.

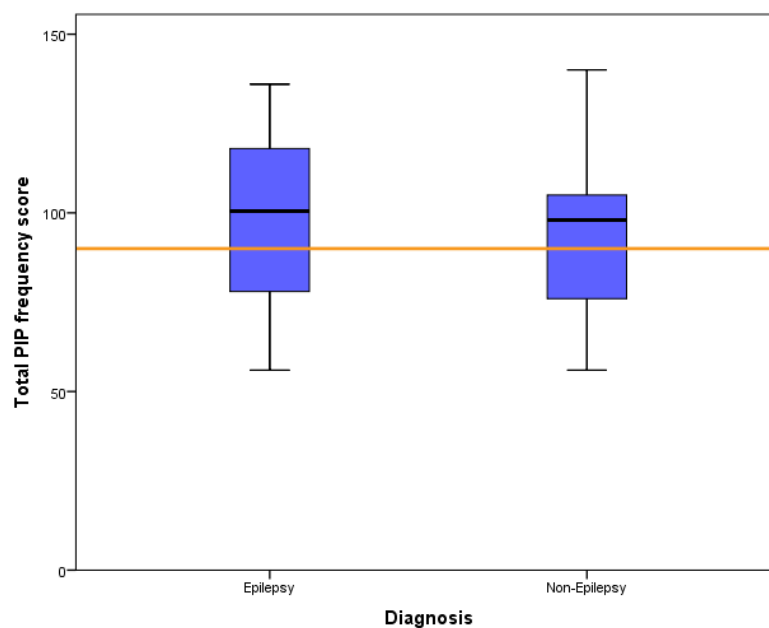


Figure 18: Box plot graph of Total PIP-f scores between cohorts at T0

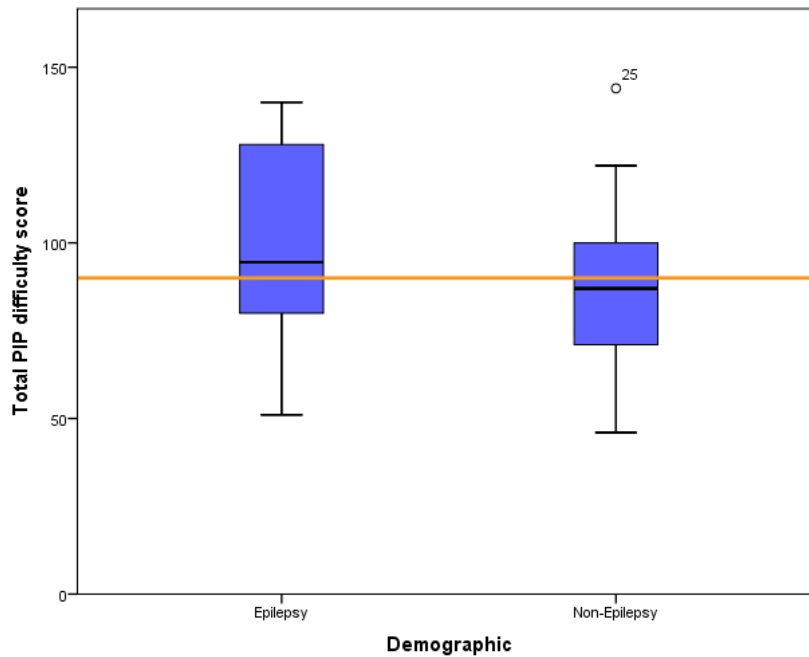


Figure 19: Box plot graph of total PIP-d scores between cohorts at T0

Following the graphical representation of this data, Table 18 & 19, an overlap was apparent between the ranges and inter-quartile ranges of the 2 cohorts. Statistical analysis was carried out using the Mann-Whitney U test to identify if there were any significant differences between the epilepsy and non-epilepsy cohorts. As demonstrated by the calculations below no significant difference was found between the stress levels between the cohorts at time of presentation.

Hypothesis Test Summary				
	Null Hypothesis	Test	Sig.	Decision
1	The medians of total_PIPfreq are the same across categories of Demographic.	Independent-Samples Median Test	.863	Retain the null hypothesis.
2	The distribution of total_PIPfreq is the same across categories of Demographic.	Independent-Samples Mann-Whitney U Test	.563	Retain the null hypothesis.
3	The medians of total_PIPdiff are the same across categories of Demographic.	Independent-Samples Median Test	.863	Retain the null hypothesis.
4	The distribution of total_PIPdiff is the same across categories of Demographic.	Independent-Samples Mann-Whitney U Test	.278	Retain the null hypothesis.

Asymptotic significances are displayed. The significance level is .05.

Figure 20: Mann-Whitney U results comparing the PIP-f and PIP-d scores between Epilepsy and Non-Epilepsy cohorts at T0

Comparison between cohorts was then completed for the T6 data, detailed in the box plot graphs below. It was immediately apparent that the distribution of the scores was not only different to that at T0 but also different between the cohorts. To confirm this observation statistical testing was conducted and, as illustrated in Figure 23, significantly higher stress scores were reported by carers of the epilepsy cohorts compared to the non-epilepsy group following diagnosis.

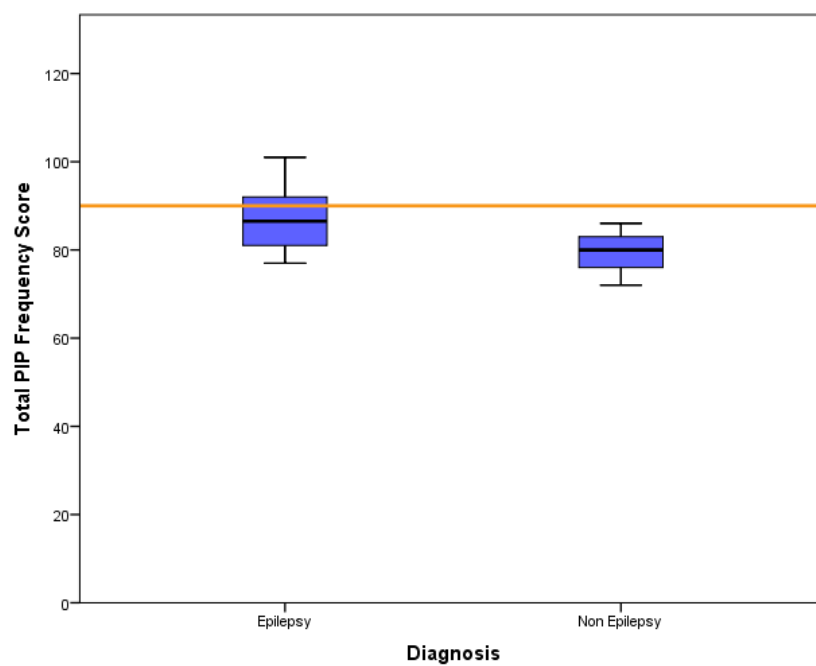


Figure 21: Box plot of PIP-f scores between cohorts at T6

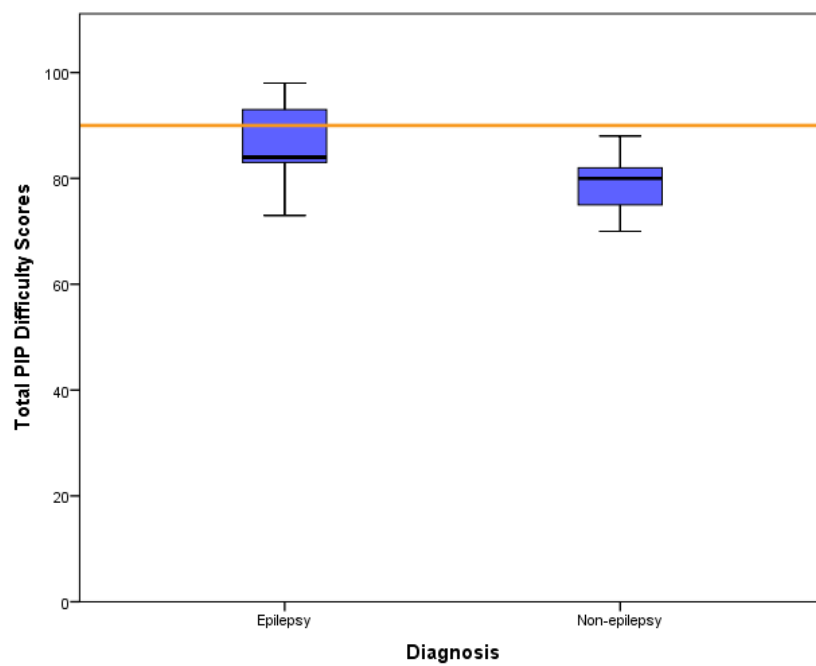


Figure 22: Box plot graph of PIP-d scores between cohorts at T6

Hypothesis Test Summary				
	Null Hypothesis	Test	Sig.	Decision
1	The medians of tot_PIP_F are the same across categories of Demographic.	Independent-Samples Median Test	.018	Reject the null hypothesis.
2	The distribution of tot_PIP_F is the same across categories of Demographic.	Independent-Samples Mann-Whitney U Test	.001	Reject the null hypothesis.
3	The medians of tot_PIP_D are the same across categories of Demographic.	Independent-Samples Median Test	.001	Reject the null hypothesis.
4	The distribution of tot_PIP_D is the same across categories of Demographic.	Independent-Samples Mann-Whitney U Test	.003	Reject the null hypothesis.

Asymptotic significances are displayed. The significance level is .05.

Figure 23: Mann Whitney U results comparing PIP-f and PIP-d scores between Epilepsy and Non-Epilepsy cohorts at T6

Descriptive Analysis of Individual Subscales

Descriptive statistics for each of the subscale scores were calculated for the overall populations' data, illustrated in Table 20. These showed that medical care demonstrated the highest mean and median compared to the other frequency subscales. Of the difficulty subscales, emotional distress was most prevalent, as suggested by mean and median values of 39.44 and 38 respectively.

Table 20: Descriptive Statistics Summary of PIP subscales in the total sample population at T0 (n=39)

	Total PIP-f	Communication Frequency	Medical care Frequency	Emotional Distress Frequency	Role Function Frequency	Total PIP-d	Communication Difficulty score	Medical Care Difficulty	Emotional Distress Difficulty	Role Function Difficulty
Mean	96.13	19.31	18.18	38.79	20.18	92.62	16.87	15.28	39.44	19.95
Median	98.00	19.00	18.00	39.00	21.00	91.00	16.00	14.00	38.00	19.00
Std. Deviation	23.036	5.277	5.180	10.191	6.692	25.952	6.071	5.341	12.592	7.330
Range	84	23	21	47	24	98	27	21	51	28
25	76.00	15.00	14.00	31.00	14.00	76.00	13.00	11.00	34.00	14.00
50	98.00	19.00	18.00	39.00	21.00	91.00	16.00	14.00	38.00	19.00
75	115.00	22.00	22.00	44.00	24.00	109.00	20.00	18.00	46.00	24.00

5.6.5 Semi-Structured Stress Questionnaire Analysis

The SSSQ was the second measure of stress utilised by this study. The histograms below, Figures 24 & 25, illustrate the range of scores reported for the SSSQ for both the T0 and T6 data. The reference lines on the x axis at x=10 indicate the suggested cut-off value for evaluating whether or not a carer is experiencing greater levels of stress.

The overall range at T0 was between 2 and 69 whilst that at T6 spanned between 4 and 70. Interestingly, using the advised boundary score of 10, at T0 *i.e.* time of presentation 79% of carers were documenting increased stress levels compared to 76% at time of diagnosis.

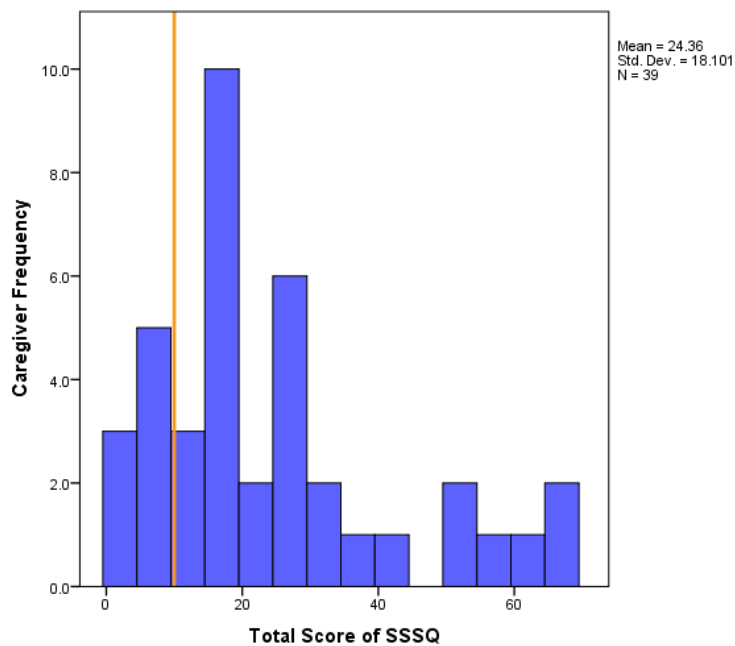


Figure 24: Histogram of total scores for the SSSQ in the total sample at T0

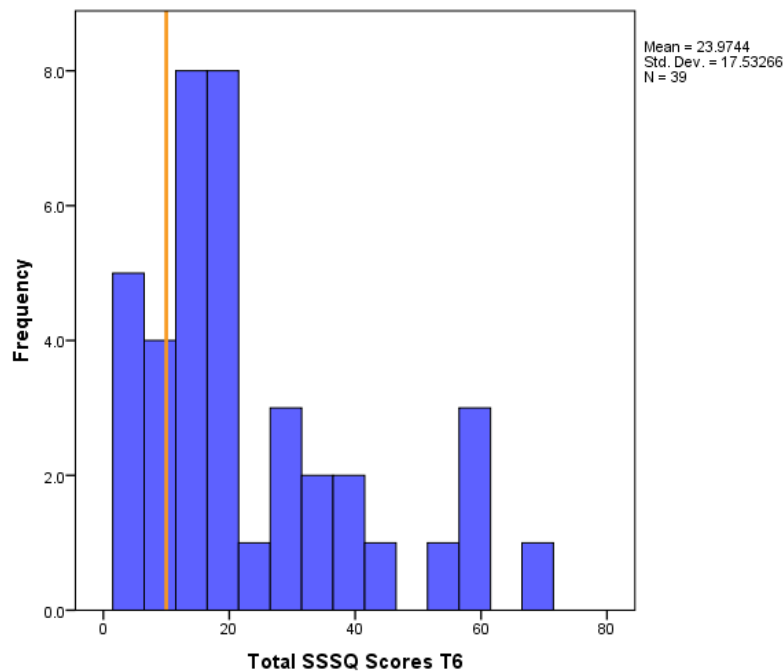


Figure 25: Histogram of total scores for the SSSQ in the total sample at T6

Total SSSQ data was also analysed between cohorts for any statistically significant difference at T0 and T6 time points. The potential of skewed data necessitates the box plot graph illustrated in Figures 26 and 28. Three outliers on these graphs are numbered and indicated by circle points.

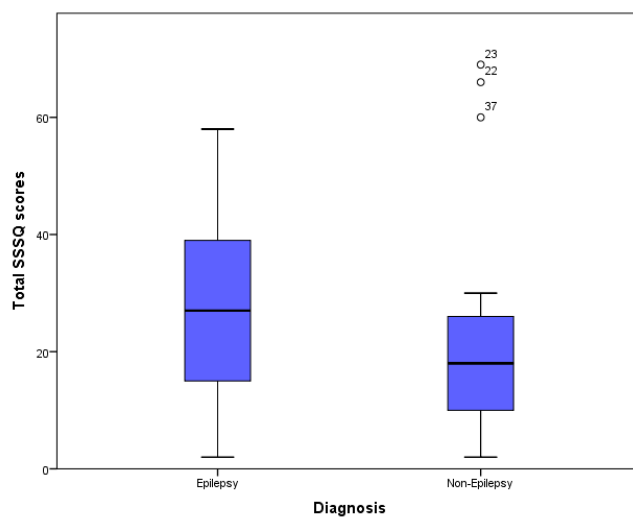


Figure 26: Box plot graph of total SSSQ scores between cohorts at T0

Results of the Mann-Whitney U test, shown below, indicate that at initial presentation there was no statistically significant difference between the SSSQ scores of the Epilepsy and Non-Epilepsy Cohorts

Hypothesis Test Summary				
	Null Hypothesis	Test	Sig.	Decision
1	The medians of max 96 are the same across categories of Demographic.	Independent-Samples Median Test	.266	Retain the null hypothesis.
2	The distribution of max 96 is the same across categories of Demographic.	Independent-Samples Mann-Whitney U Test	.242	Retain the null hypothesis.

Asymptotic significances are displayed. The significance level is .05.

Figure 27: Mann Whitney U test results for difference between SSSQ scores between epilepsy cohorts at T0

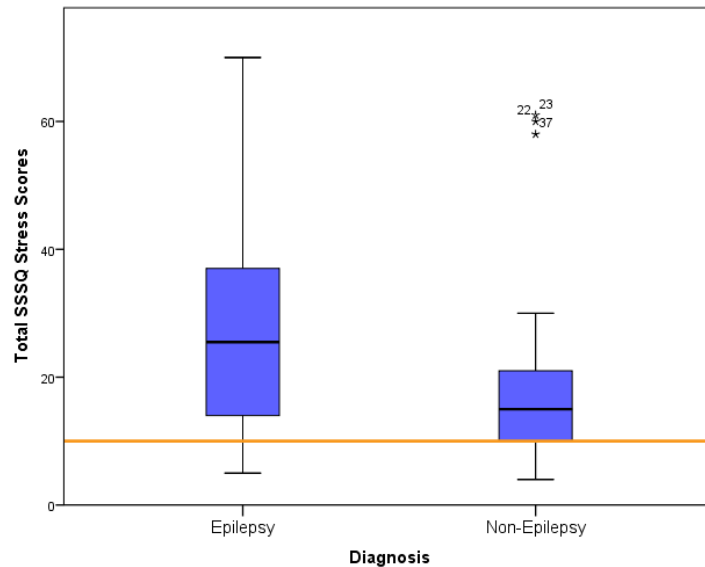


Figure 28: Box plot graph of total SSSQ scores between the cohorts at T6

Hypothesis Test Summary				
	Null Hypothesis	Test	Sig.	Decision
1	The medians of Total_SSQ_Stress_6 are the same across categories of Demographic.	Independent-Samples Median Test	.040	Reject the null hypothesis.
2	The distribution of Total_SSQ_Stress_6 is the same across categories of Demographic.	Independent-Samples Mann-Whitney U Test	.048	Reject the null hypothesis.

Asymptotic significances are displayed. The significance level is .05.

Figure 29: Mann-Whitney U test results of statistically significant differences between cohorts at T6

For the SSSQ data at T6 it is apparent from the box plot graph that the inter-quartile ranges of both cohorts lie above the score of 10 (indicator for increased stress levels). The distributions of the different cohorts were also observed to be different and this was confirmed by Mann-Whitney U testing, illustrated above. It shows that SSSQ scores at time of diagnosis were significantly higher in the epilepsy cohort compared to the non-epilepsy.

5.6.6 General Health Questionnaire-28 Analysis

The GHQ is used to measure the variable carer health and focuses predominantly on mental aspects of health. Using the bimodal scoring method (0,0,1,1) for this questionnaire as discussed in Chapter 4, the maximum total score for this tool is 18.

As shown in the histogram below, the range of scores across the entire sample was from 0 to 14 with the mode being 0.

Also seen below are the box plot graphs describing the distributions between the cohorts.

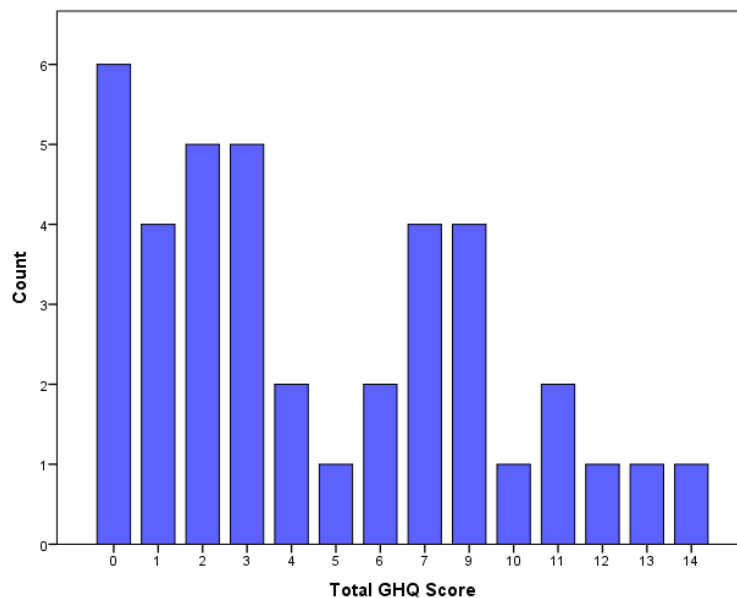


Figure 30: Histogram of overall study population total GHQ scores at T0

The distribution of total GHQ scores across the entire sample of carers shows a wide range. Although the overall mode was 0 and the median score was 3 it is still interesting to note that 36% of primary caregivers at T0 were reporting scores greater than 5- which is the suggested value by Goldberg for indication of psychological distress.

Descriptive statistics for the individual subscales forming the GHQ were conducted to determine whether some aspects of health had greater frequency than others.

Table 21: Descriptive statistics for total and subscale GHQ scores at T0

Descriptive Statistics					
	General health questionnaire somatic symptoms	General health questionnaire anxiety and insomnia	General health questionnaire social dysfunction	General health questionnaire depression	Total GHQ Scores
Mean	1.79	1.64	1.08	.36	4.85
Median	2.00	2.00	.00	.00	3.00
Mode	0	0	0	0	0
Range	4	4	4	3	14
Percentiles 25	.00	.00	.00	.00	1.00
Percentiles 50	2.00	2.00	.00	.00	3.00
Percentiles 75	3.00	3.00	2.00	1.00	9.00

As seen in Table 21 there were minimal difference between the GHQ subscales, although the means and medians for the somatic symptoms and anxiety and insomnia scales were higher than the social dysfunction and depression scales, 1.79 and 1.64 compared to 1.08 and 0.36.

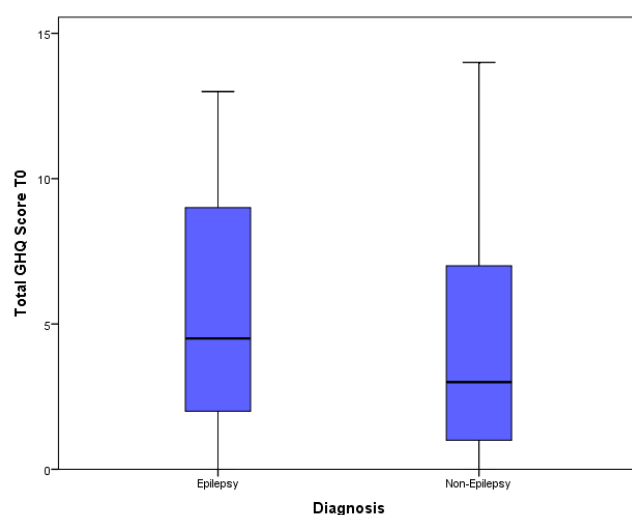


Figure 31: Box plot graph of total GHQ scores between the cohorts at T0

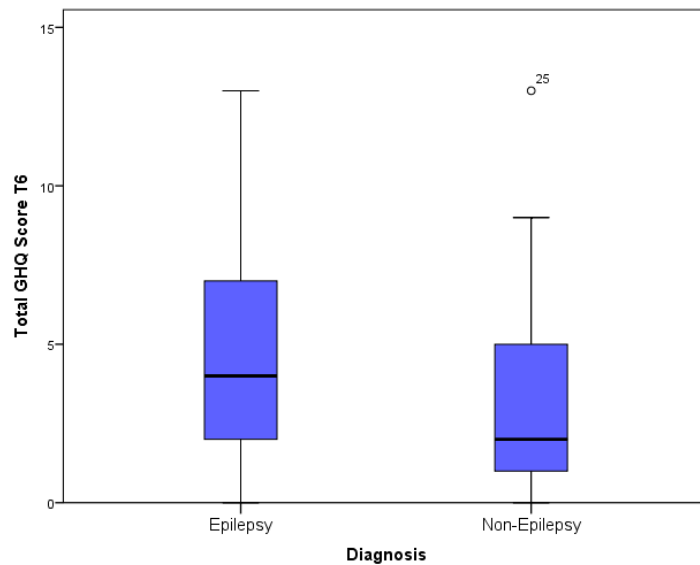


Figure 32: Box plot graph of total GHQ scores between the cohorts at T6

On separating the total GHQ scores by cohort it was evident that both the epilepsy and non-epilepsy carers demonstrated a similar range scores, Figures 31 and 32. Despite the epilepsy cohort having a slightly greater median and wider inter-quartile range upon statistical testing it was determined that there was no significant difference between the two on the basis of the GHQ scores, illustrated in Figure 33 below.

Hypothesis Test Summary				
	Null Hypothesis	Test	Sig.	Decision
1	The medians of Total GHQ Score T6 are the same across categories of Epilepsy Status.	Independent-Samples Median Test	.167	Retain the null hypothesis.
2	The distribution of Total GHQ Score T6 is the same across categories of Epilepsy Status.	Independent-Samples Mann-Whitney U Test	.239	Retain the null hypothesis.

Asymptotic significances are displayed. The significance level is .05.

Figure 33: Mann Whitney U test results for T6 total GHQ scores

Comparative analysis between the cohorts of their total GHQ scores at T6 revealed overlapping ranges and inter-quartile ranges with only 2 points between the median values of the two groups. Statistical analysis indicated that as at time of presentation there were also no significant differences between the cohorts at time of diagnosis.

5.6.7 Rotter's Locus of Control Analysis

Rotter's locus of control questionnaire aims to differentiate between those with an internal and those with an external locus of control. Using the same methods as in previous studies the median value of the total group will be used as the cut-off score to determine which carers have predominantly internal or external loci of control. Scores at either extreme of this scale are deemed to be detrimental (Beretvas et al., 2008). 39 carers completed this questionnaire at both time points. The histogram below, Figure 34, indicates the distribution of their scores with the orange reference line on the x axis indicating a balanced score. The range for the overall sample, as illustrated in the graph, spanned from 7 to 20.

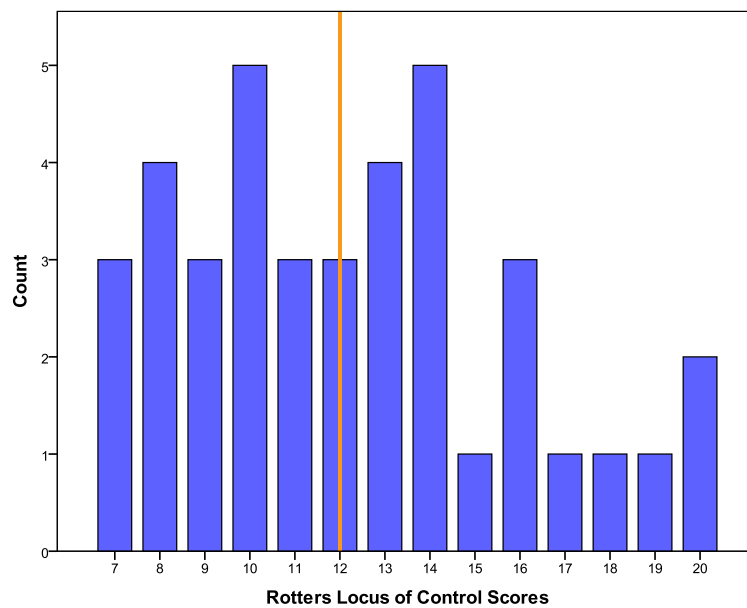


Figure 34: Histogram for total sample populations Rotter's LoC scores at T0

In addition to examining the overall spread of scores, the distributions of the separate cohorts at time points T0 and T6 are illustrated in the following box plot chart, Figure 35. Reference lines have been placed on the y axes at a score of 12 indicating a balanced internal-external LoC

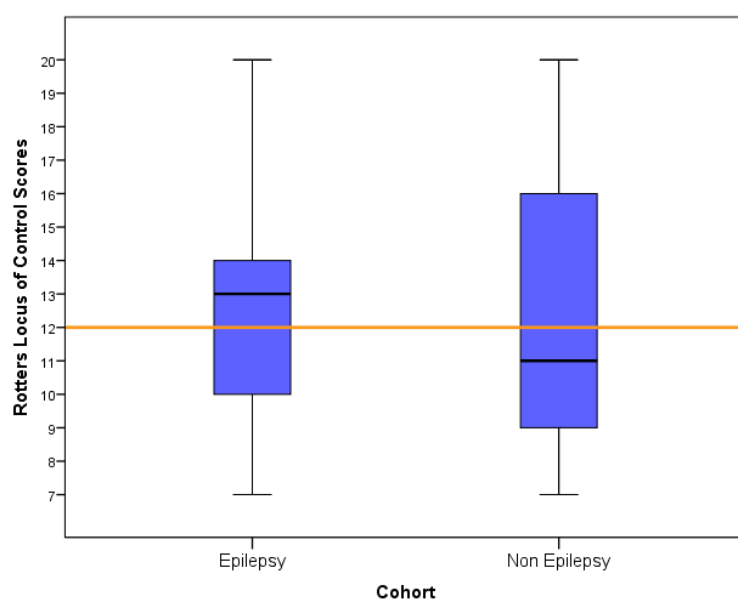


Figure 35: Box plot graph of Rotter's LoC scores between cohorts at T0

Although the means for the epilepsy and non epilepsy cohorts fell above and below the median, seen in Figure 35 above, they exhibited similar ranges and distributions and no statistically significant difference found.

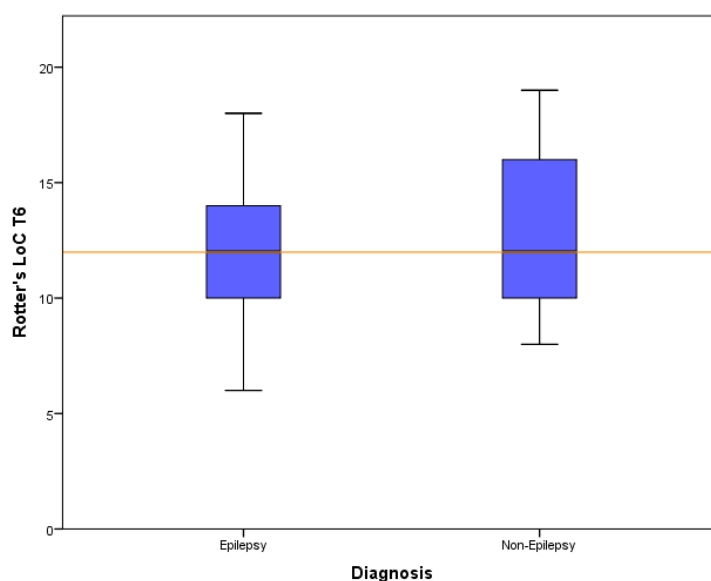


Figure 36: Box plot graph of Rotter's LoC scores between cohorts at T6

At T6 the overall median value for the carers was again 12 and, upon segregation into groups the median values for both cohorts were also 12. This, in conjunction with similar inter-quartile ranges suggested there was no difference between the cohorts. This assertion was then confirmed via statistical analyses, which indicates that there was no significant difference in carer health scores between the epilepsy and non-epilepsy cohorts.

5.6.8 Brief COPE Analysis

Unlike the other questionnaires the COPE does not produce a total score but instead aims to gather information on range of different mechanisms used. Individual scores from each of the subscales are produced and summation of certain predefined subscales provides composite scores for problem focused, emotion focused and dysfunction focused coping.

Descriptive statistics, as documented below, were conducted for each of the individual subscales to determine if there was a particularly prevalent coping strategy being used by this sample of carers.

Table 22 shows how carers identified with some coping mechanism mechanisms more than others. Acceptance, a positive coping strategy was found to have the highest mean and median value, 5.79 and 6 respectively. This is in contrast to the substance misuse coping method which showed a mean of 2.36 and median 2. Additionally while the range of response for the majority of the individual subscales was 6 the self-distraction strategy had the maximum range of 10 compared to those of 4 for substance misuse and behavioural disengagement.

Table 22: Descriptive statistics for COPE subscales at T0

Descriptive Statistics							
	Coping strategies self distraction	Coping strategies active distraction	Coping strategies denial	Coping strategies substance use	coping strategies emotional support	coping strategies instrumental support	coping strategies behavioural disengagement
Mean	4.00	5.18	2.51	2.36	4.85	5.21	2.51
Median	3.00	5.00	2.00	2.00	5.00	6.00	2.00
Mode	2	4 ^a	2	2	2 ^a	6	2
Range	10	6	6	4	6	6	4
Percentile 25	2.00	4.00	2.00	2.00	3.00	3.00	2.00
Percentile 50	3.00	5.00	2.00	2.00	5.00	6.00	2.00
Percentile 75	5.00	7.00	2.00	2.00	7.00	7.00	2.00

a. Multiple modes exist. The smallest value is shown

Descriptive Statistics cont.

	coping strategies venting	coping strategies positive reframing	coping strategies planning	coping strategies humour	coping strategies acceptance	coping strategies religion	coping strategies self blame
Mean	3.08	3.77	4.74	2.85	5.79	3.23	3.23
Median	2.00	3.00	5.00	2.00	6.00	2.00	2.00
Mode	2	2 ^a	3	2	8	2	2
Range	6	5	6	6	6	6	6
Percentile 25	2.00	2.00	3.00	2.00	4.00	2.00	2.00
Percentile 50	2.00	3.00	5.00	2.00	6.00	2.00	2.00
Percentile 75	4.00	5.00	6.00	3.00	8.00	4.00	5.00

a. Multiple modes exist. The smallest value is shown

In addition, the 3 composite scales analysed to ascertain whether there were any broad differences in coping between the carers of children with and without epilepsy. The box plot graphs below represent the data collected at T0 *i.e.* at time of initial presentation. From inspection of these it was unclear if there was any difference. However, the following statistical analyses recorded p values >0.05, therefore indicating that there was no significant difference between the cohorts based upon their scores of problem focused, emotion focused and dysfunctional coping.

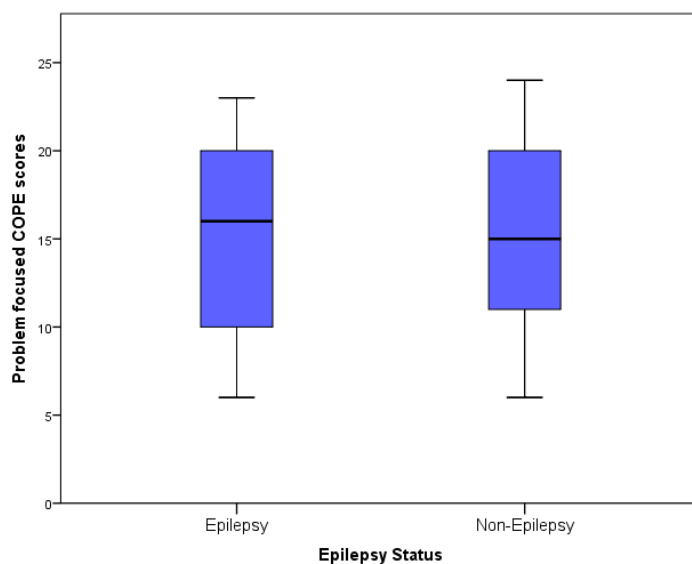


Figure 37: Problem focused coping scores between cohorts at T0

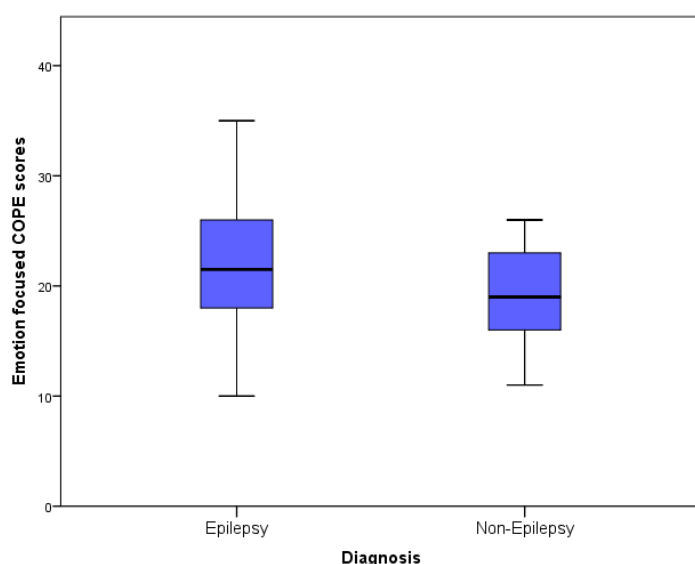


Figure 38: Emotion focused coping scores between cohorts at T0

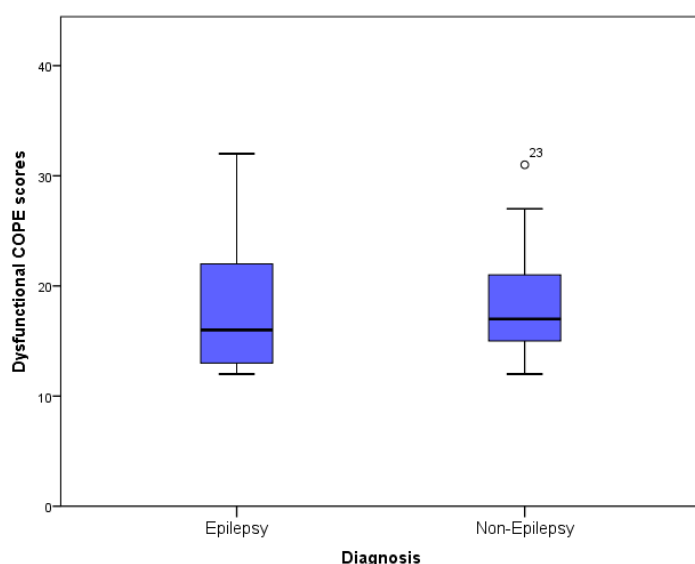


Figure 39: Dysfunctional coping scores between cohorts at T0

Separation of the 3 aggregate coping scores based on epilepsy status showed that for problem focused coping methods both cohorts had a similar distribution, although the median value for the epilepsy group was marginally higher than that of the non-epilepsy. For the emotion focused scores however, although the size of the inter-quartile range was equal, carers of the epilepsy contingent demonstrated a broader range of scores with a higher median than that of non epilepsy. Finally, on analysis of the dysfunctional coping scores, the epilepsy cohort again had a wider overall distribution of scores although for this scale the non-epilepsy group had a greater median value

However, as demonstrated by Figure 40 below, these differences were not found to be significant.

Hypothesis Test Summary				
	Null Hypothesis	Test	Sig.	Decision
1	The medians of Problem focused coping are the same across categories of Epilepsy Status.	Independent-Samples Median Test	.863	Retain the null hypothesis.
2	The distribution of Problem focused coping is the same across categories of Epilepsy Status.	Independent-Samples Mann-Whitney U Test	.977	Retain the null hypothesis.
3	The medians of Emotion focused coping are the same across categories of Epilepsy Status.	Independent-Samples Median Test	.639	Retain the null hypothesis.
4	The distribution of Emotion focused coping is the same across categories of Epilepsy Status.	Independent-Samples Mann-Whitney U Test	.175	Retain the null hypothesis.
5	The medians of Dysfunctional coping are the same across categories of Epilepsy Status.	Independent-Samples Median Test	.563	Retain the null hypothesis.
6	The distribution of Dysfunctional coping is the same across categories of Epilepsy Status.	Independent-Samples Mann-Whitney U Test	.561	Retain the null hypothesis.

Asymptotic significances are displayed. The significance level is .05.

Figure 40: Mann-Whitney U results for differences in coping methods between cohorts at T0

A similar procedure was conducted for the T6 data. On this occasion Mann-Whitney U testing found that there was a significant difference between the cohorts for the dysfunctional coping scale, as evidenced by $p=0.40$ seen in Figure 41. No significant differences however were found on the problem focused and emotion focused composite scales.

Hypothesis Test Summary				
	Null Hypothesis	Test	Sig.	Decision
1	The medians of Problem focused coping T6 are the same across categories of Epilepsy Status.	Independent-Samples Median Test	.863	Retain the null hypothesis.
2	The distribution of Problem focused coping T6 is the same across categories of Epilepsy Status.	Independent-Samples Mann-Whitney U Test	.471	Retain the null hypothesis.
3	The medians of Emotion focused coping T6 are the same across categories of Epilepsy Status.	Independent-Samples Median Test	.863	Retain the null hypothesis.
4	The distribution of Emotion focused coping T6 is the same across categories of Epilepsy Status.	Independent-Samples Mann-Whitney U Test	.746	Retain the null hypothesis.
5	The medians of Dysfunctional coping T6 are the same across categories of Epilepsy Status.	Independent-Samples Median Test	.040	Reject the null hypothesis.
6	The distribution of Dysfunctional coping T6 is the same across categories of Epilepsy Status.	Independent-Samples Mann-Whitney U Test	.003	Reject the null hypothesis.

Asymptotic significances are displayed. The significance level is .05.

Figure 41: Mann-Whitney U test results for differences in coping methods between cohorts at T6

5.6.9 Family Support Scale Analysis

The FSS seeks to measure the amount of support received by an individual. It produces an overall score representing the total amount of support obtained, in addition to subscales which seek to investigate the helpfulness of the different potential sources of support available to the person.

Total Levels of Support

Firstly, the total level of support score was analysed for the whole sample population at T0. A large spread of data was found, as illustrated in Figure 42, with a range between 20 and 62 (the maximum possible score is 90)

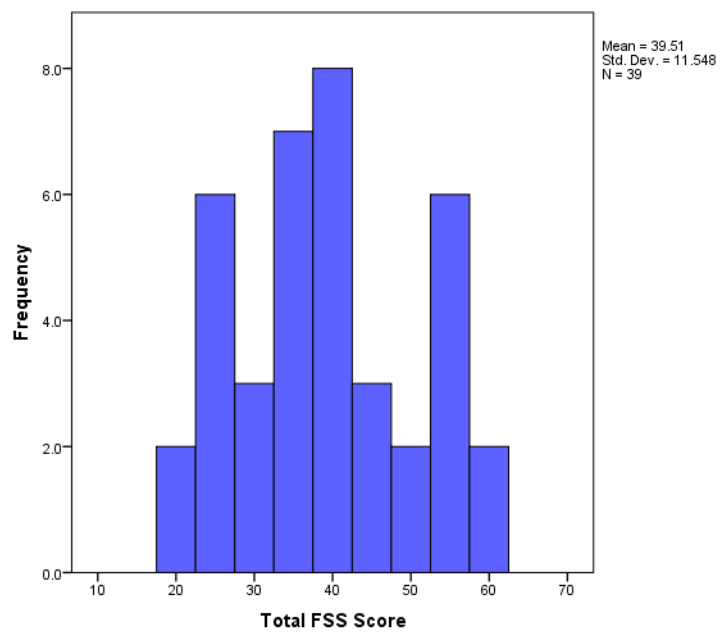


Figure 42: Histogram showing total sample population scores for total FSS scores at T0

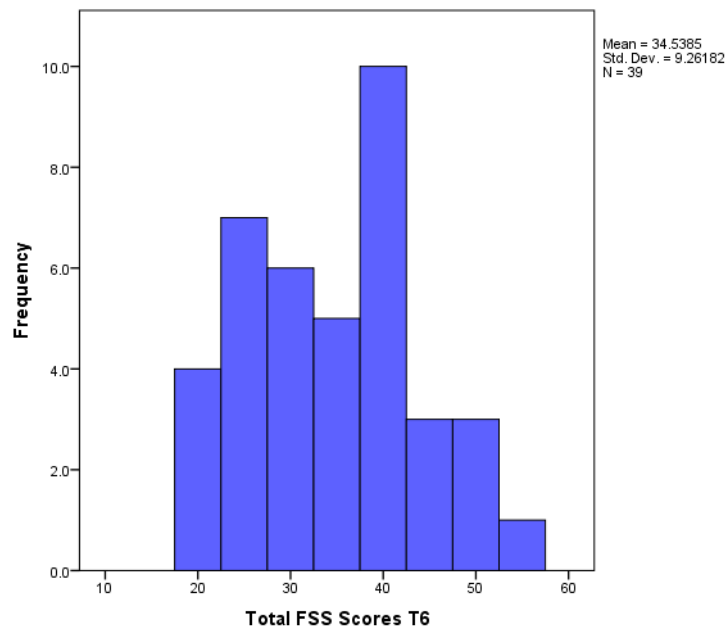


Figure 43: Histogram showing total sample population scores for total FSS scores at T6

The histograms above, Figures 42 and 43, indicate the spread of FSS scores at both timepoints. They exhibit a distribution similar to a Gaussian. While the scores at T0 had a slightly larger range, T6 results fell closer together. Additionally the T0 mean, 39.51 was larger compared to the T6 mean of 34.5

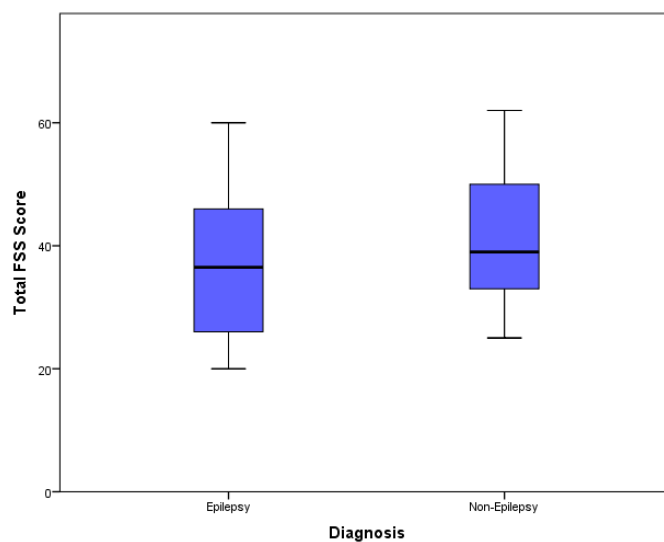


Figure 44: Box plot graph of total FSS scores between cohorts at T0

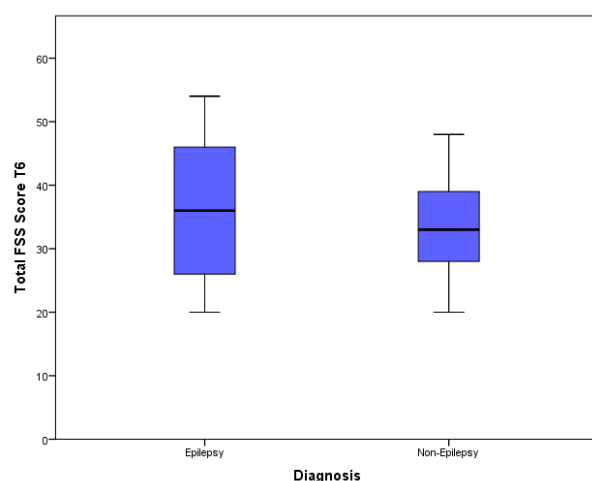


Figure 45: Box plot graph of total FSS scores between cohorts at T6

The box plot graphs above illustrate that there was no significant difference between the cohorts total levels of received support at both time of presentation and time of diagnosis 6 months later. This was further confirmed by Mann-Whitney U testing, results of which are displayed below in Figure 46.

Hypothesis Test Summary				
	Null Hypothesis	Test	Sig.	Decision
1	The medians of Total FSS Scores are the same across categories of Epilepsy Status.	Independent-Samples Median Test	.823	Retain the null hypothesis.
2	The distribution of Total FSS Scores is the same across categories of Epilepsy Status.	Independent-Samples Mann-Whitney U Test	.236	Retain the null hypothesis.
3	The medians of Total FSS T6 are the same across categories of Epilepsy Status.	Independent-Samples Median Test	.863	Retain the null hypothesis.
4	The distribution of Total FSS T6 is the same across categories of Epilepsy Status.	Independent-Samples Mann-Whitney U Test	.499	Retain the null hypothesis.

Asymptotic significances are displayed. The significance level is .05.

Figure 46: Mann-Whitney U tests for T0 and T6 comparisons of FSS scores between cohorts

Table 23: Descriptive statistics for FSS subscales in the total sample population

Descriptive Statistics						
	N	Range	Minimum	Maximum	Mean	Std. Deviation
FSS partner support	39	15	0	15	9.03	4.319
FSS informal kinship	39	24	0	24	11.31	5.449
FSS formal kinship	39	11	0	11	6.15	2.681
FSS social organisation	39	11	1	12	6.00	2.974
FSS professional services	39	9	3	12	7.03	2.560
Total FSS	39	42	20	62	39.51	11.548

Valid N (listwise)	39					
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Hypothesis Test Summary				
	Null Hypothesis	Test	Sig.	Decision
1	The distribution of fss partner support is the same across categories of Epilepsy Status.	Independent-Samples Mann-Whitney U Test	.046	Reject the null hypothesis.
2	The distribution of fss informal kinship is the same across categories of Epilepsy Status.	Independent-Samples Mann-Whitney U Test	.854	Retain the null hypothesis.
3	The distribution of fss formal kinship is the same across categories of Epilepsy Status.	Independent-Samples Mann-Whitney U Test	.630	Retain the null hypothesis.
4	The distribution of fss social organisation is the same across categories of Epilepsy Status.	Independent-Samples Mann-Whitney U Test	.640	Retain the null hypothesis.
5	The distribution of fss professional services is the same across categories of Epilepsy Status.	Independent-Samples Mann-Whitney U Test	.262	Retain the null hypothesis.

Asymptotic significances are displayed. The significance level is .05.

Figure 47: Mann-Whitney U testing of FSS subscale differences between cohorts at T6

Statistical testing was conducted on the individual elements present within the FSS, seen in Table 21. It was of interest to investigate whether different sources of support were more prevalent between the carers of children with epilepsy and those without. No significant findings were present for the T0 data. However, at T6 the levels of partner support were significantly higher in the epilepsy cohort than that of the non-epilepsy. This is illustrated in Figure 47 above (N.B partner support is not a formal subscale of the FSS).

5.6.10 Family Needs Survey Analysis

As with the previous questionnaires, 39 carers completed the FNS at both time points in the study. The scoring method used was the 0, 0, 1 technique in order to highlight definite needs. This means that the maximum possible total score for this questionnaire is 35.

The FNS was first broken down into its component subscales to observe whether there was a particular area in which all families identified a strong need. This is illustrated by Table 24 below. It shows that the counselling needs were exhibited by very few carers as the mean median and mode were all around zero whereas descriptive statistics for the remaining subscales were greater.

Table 24: Descriptive statistics for subscales of the FNS

Descriptive Statistics				
	Family needs survey resource need	Family needs survey information need	family needs survey counselling need	Total FNS Score
Mean	3.08	2.33	.31	5.7179
Median	2.00	3.00	.00	5.0000
Mode	0	0	0	.00
Range	10	6	3	17.00
Percentiles				
25	.00	.00	.00	.0000
50	2.00	3.00	.00	5.0000
75	6.00	4.00	.00	10.0000

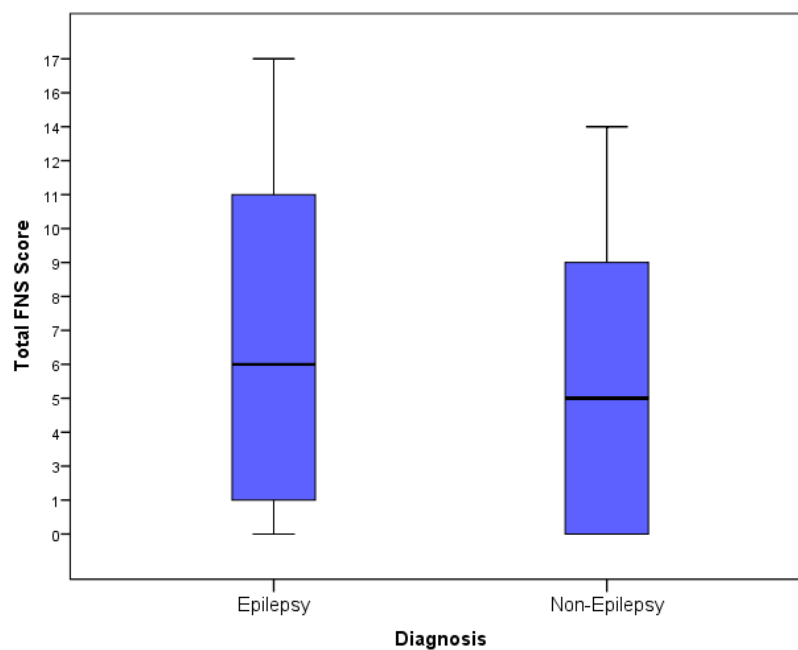


Figure 48: Box plot graph of total FNS scores between the cohorts at T0

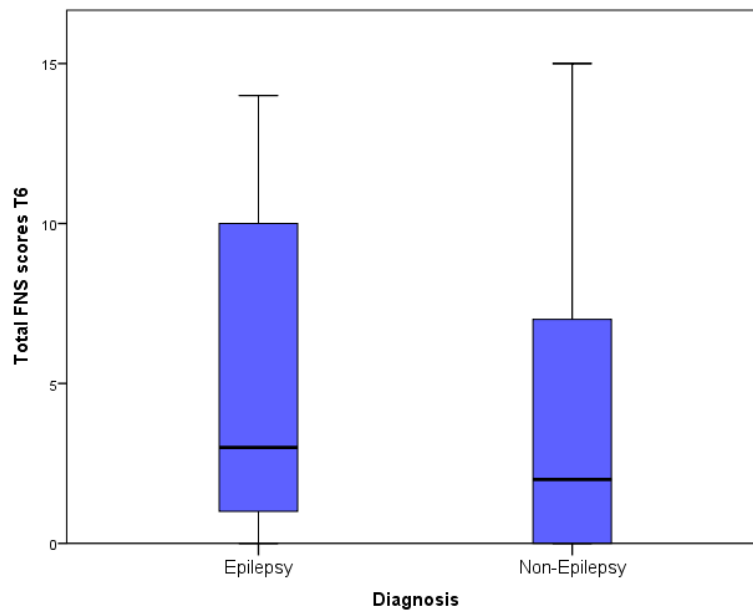


Figure 49: Box plot graph of total FNS scores between the cohorts at T6

The box plot graphs above, Figures 48 & 49, indicate that at time of presentation all carers exhibited a range of total needs scores although the median value for the epilepsy cohort was 6 compared to 5 in that of the non-epilepsy. Following this at T6 *i.e.* time of diagnosis, although a wide range was still present, medians of both cohorts fell to 3 and 2 respectively.

5.6.11 Family Adaptability and Cohesion Scale IV analysis

This questionnaire produces scales to help quantify overall family function as well as family communication and family satisfaction. For this subsection of the results n=38 due to one missing data field in a questionnaire of a caregiver at T6.

The total circumplex ratio was the first aspect of the FACES tool to be analysed. This is purported to represent an overall view of family functioning.

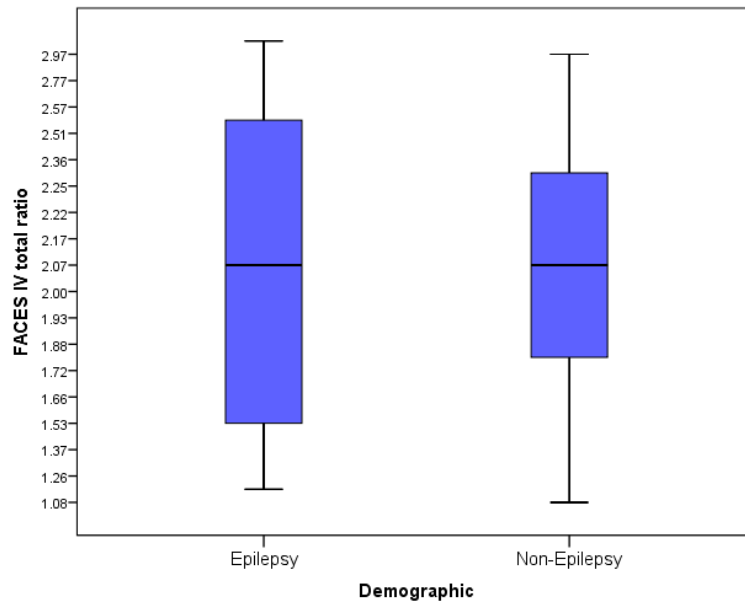


Figure 50: Comparison of total circumplex ratio scores between cohorts at T0

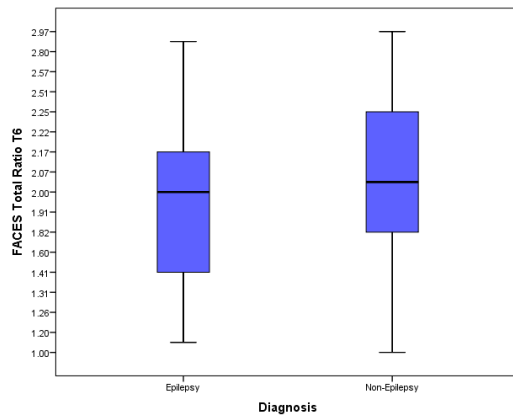


Figure 51: Box plot graph of FACES IV total ratio between cohorts at T6

In addition to the total circumplex ratio, it was also identified via the literature study and personal communications with Dr Andrew Curran that the family communication and family satisfaction scales produced by the FACES tool were also areas of particular interest. Therefore, comparisons between the cohorts of these two scales were carried out both at T0 and T6 time points.

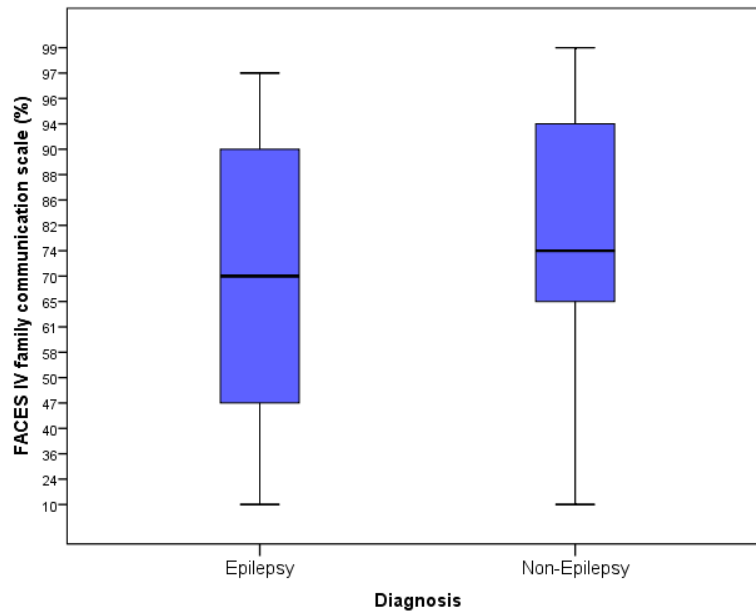


Figure 52: Box plot of FACES IV family communications scale scores at T0

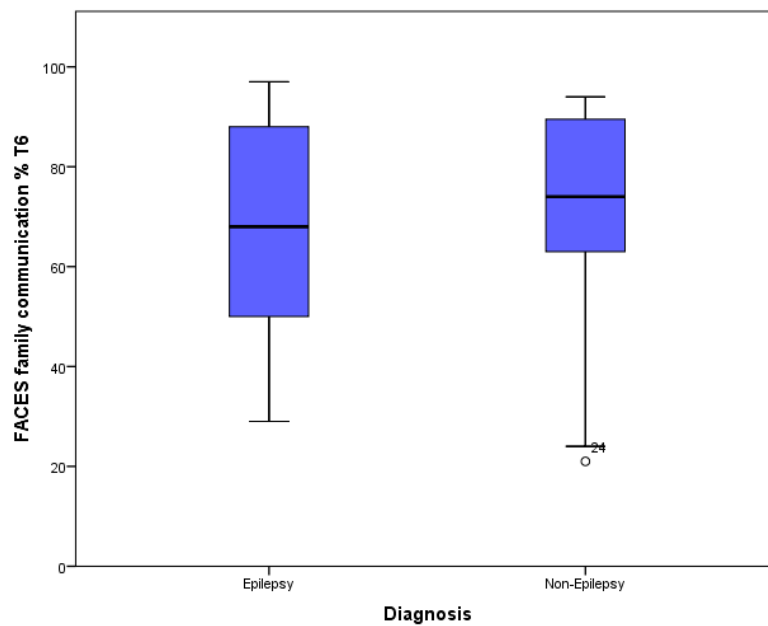


Figure 53: Box plot of FACES IV family communications score at T6

For the family communications scale, although the overall ranges between the groups were very wide, the inter-quartile range of the non-epilepsy group was observed to be smaller at both time points. This is shown by Figure 52 and 53. The median values changed from 70 and 73 at T0 to 68 and 74 at T6 for the epilepsy and non-epilepsy cohorts respectively.

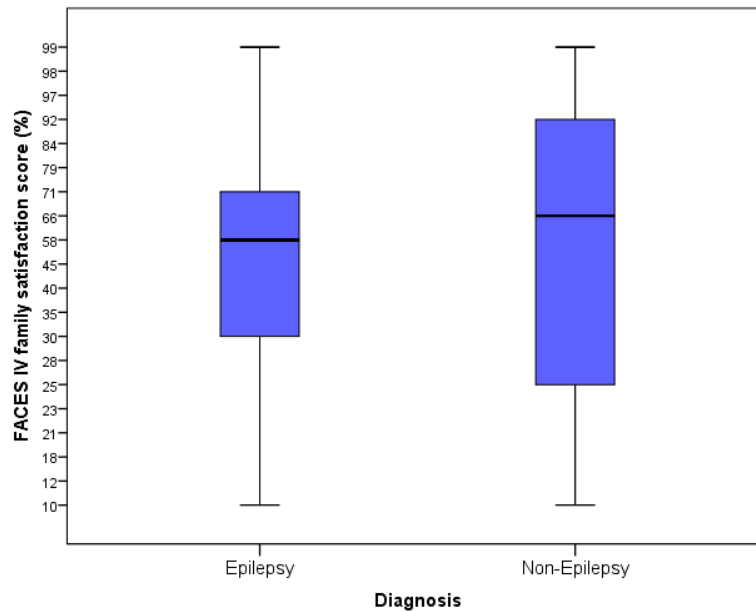


Figure 54: Box plot of FACES IV family satisfaction % scores between the two cohorts at T0

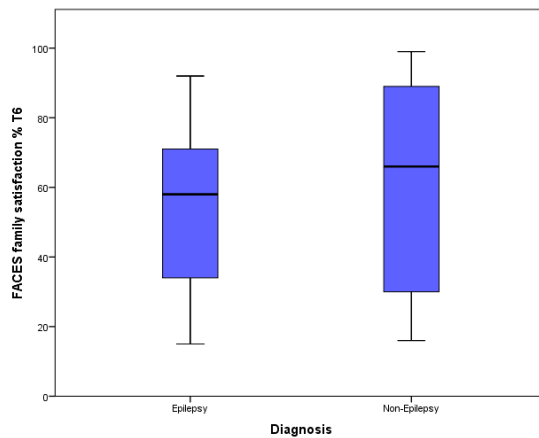


Figure 55: Box plot of FACES IV family satisfaction % between the cohorts at T6

At T6, the distribution of the satisfaction scores between the 2 cohorts was similar with the non-epilepsy cohort producing a larger inter-quartile range than the epilepsy. Despite this, the median values of the groups still fell closely, 58% compared to 66%, for the epilepsy and non-epilepsy groups respectively. This similarity was confirmed by Mann-Whitney U testing indicating that there was no significant difference between the carers family satisfaction at the time of diagnosis.

Hypothesis Test Summary				
	Null Hypothesis	Test	Sig.	Decision
1	The medians of faces family communication percentage are the same across categories of Epilepsy Status.	Independent-Samples Median Test	.921	Retain the null hypothesis.
2	The distribution of faces family communication percentage is the same across categories of Epilepsy Status.	Independent-Samples Mann-Whitney U Test	.577 ¹	Retain the null hypothesis.
3	The medians of faces family satisfaction percentage are the same across categories of Epilepsy Status.	Independent-Samples Median Test	.571	Retain the null hypothesis.
4	The distribution of faces family satisfaction percentage is the same across categories of Epilepsy Status.	Independent-Samples Mann-Whitney U Test	.596 ¹	Retain the null hypothesis.

Asymptotic significances are displayed. The significance level is .05.

¹Exact significance is displayed for this test.

Figure 56: Mann-Whitney U test results for cohort differences between FACES family communications and family satisfaction % scales at T0

Hypothesis Test Summary				
	Null Hypothesis	Test	Sig.	Decision
1	The medians of FACES family communication % T6 are the same across categories of Epilepsy Status.	Independent-Samples Median Test	.745	Retain the null hypothesis.
2	The distribution of FACES family communication % T6 is the same across categories of Epilepsy Status.	Independent-Samples Mann-Whitney U Test	.534 ¹	Retain the null hypothesis.
3	The medians of FACES family satisfaction % T6 are the same across categories of Epilepsy Status.	Independent-Samples Median Test	1.000	Retain the null hypothesis.
4	The distribution of FACES family satisfaction % T6 is the same across categories of Epilepsy Status.	Independent-Samples Mann-Whitney U Test	.486 ¹	Retain the null hypothesis.

Asymptotic significances are displayed. The significance level is .05.

¹Exact significance is displayed for this test.

Figure 57: Mann-Whitney U tests of differences between family communication and family satisfaction % between the cohorts at T6

The Figures 56 and 57 illustrate that the potential differences observed between the cohorts at both time points were not found to be statistically significant upon Mann-Whitney U testing.

5.6.12 Strengths and Difficulties Questionnaire Analysis

Total scores

Analysis of the SDQ data was begun by producing histograms of the total SDQ scores reported by the entire sample population, illustrated below in Figure 58 and 59. On each graph there are 2 reference lines on the x axis. These are placed at the scores of 14 and 17. They indicate the classifications suggested for interpreting SDQ results. A total score of 14-16 is deemed borderline whilst a score of ≥ 17 is thought to represent abnormal behavioural issues.

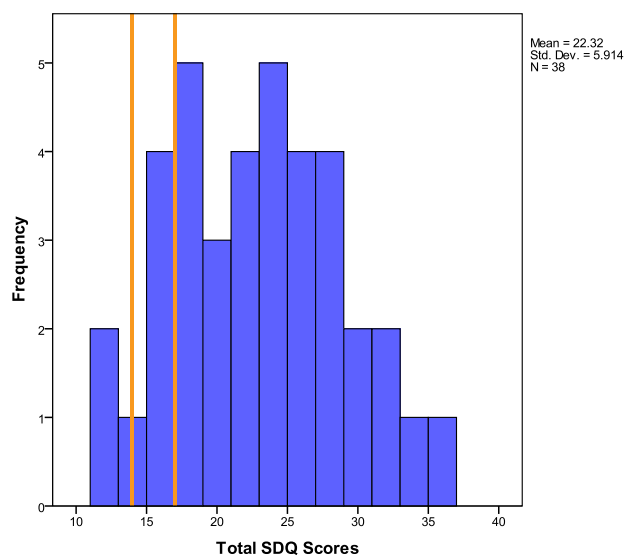


Figure 58: Histogram of Total SDQ scores for the overall sample population at T0

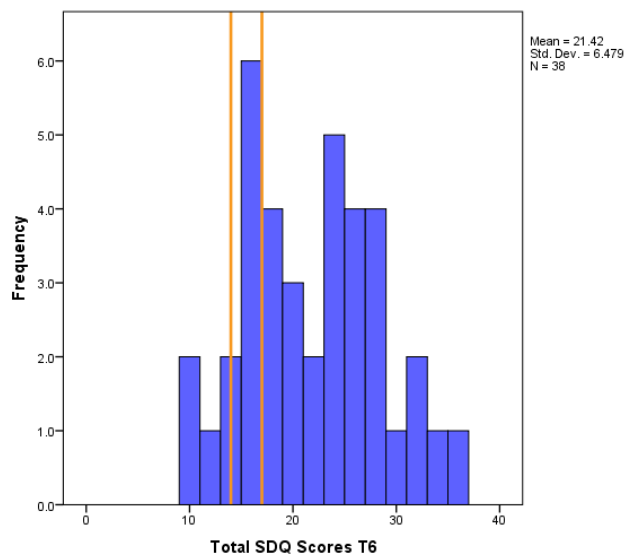


Figure 59: Histogram of total SDQ scores at T6

The distribution of total SDQ scores between T0 and T6 was fairly similar with 2 peaks in the frequency. However whilst at T0 these were at score of 18 and 24 at T6 these occurred lower, at 16 and 24-25, as seen in Figures 57 and 58.

Following analysis of the overall study population, the total SDQ scores were separated based upon cohort allocation to determine the presence of any statistical significance between the 2 groups at both time of initial presentation and following diagnosis.

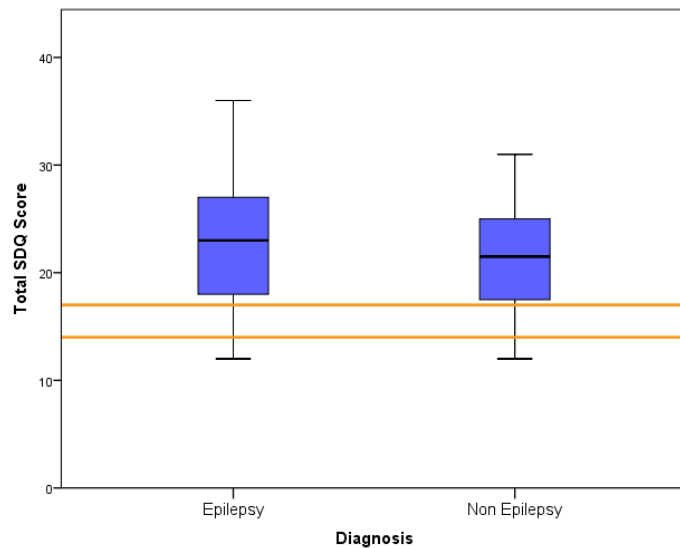


Figure 60: Box plot graph of total SDQ scores between cohorts at T0

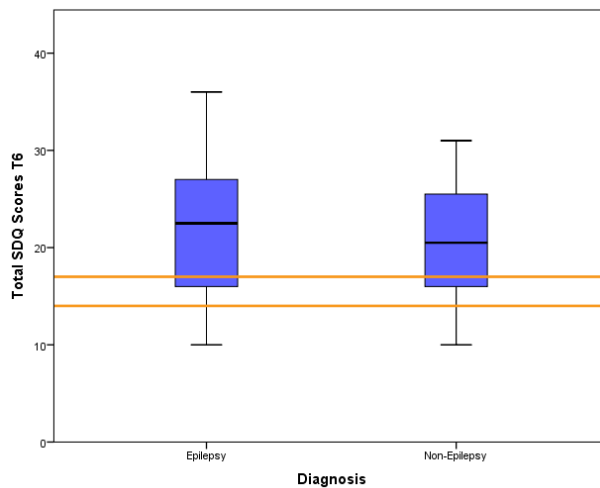


Figure 61: Box plot graph of total SDQ scores between the cohorts at T6

The box plots shown in Figures 60 and 61 illustrate that the distributions of total SDQ scores were similar between the 2 cohorts at both time points. At T6 however, the lower end of the inter-quartile ranges for both groups fell into the borderline behaviour score range when at T0 they each fell above these boundaries. The means at T6 were still outside this range, 23 and 21 for the epilepsy and non-epilepsy cohorts respectively.

Hypothesis Test Summary			
	Null Hypothesis	Test	Sig. Decision
1	The medians of Total SDQ T6 are independent- the same across categories of Epilepsy Status.	Samples Median Test	.745 Retain the null hypothesis.
2	The distribution of Total SDQ T6 is independent- the same across categories of Epilepsy Status.	Samples Mann-Whitney U Test	.784 ¹ Retain the null hypothesis.

Asymptotic significances are displayed. The significance level is .05.

¹Exact significance is displayed for this test.

Figure 62: Mann-Whitney U tests for total SDQ scores at T6

The differences in total SDQ scores between the epilepsy and non-epilepsy cohorts were not found to be significant on statistical analyses, either at T0 or at T6 as seen in Figure 62.

5.6.13 Self Image Profile Analysis

The age range of the children in the study sample and the ages for which this questionnaire is validate limits the number of datasets such that for this section of the analysis n=20. From these 12 completed the SIP-C and 8 completed the SIP-A.

Table 25: SIP scores for the total study sample

	Positive SI	Negative SI	SE
Mean	52.39	29.01	8.92
Median	54.00	32.00	4.00
Mode	47.00	16.00	6.00
Range	31.00	53.00	6.00

As seen in Table 25, overall the positive SI was much greater than the negative this is indicated by the margins between both the means, 52.39 compared to 29.01, and the median values, 54 compared to 32. Additionally the modal value for the positive SI, 47, was larger than that of the negative, 16. However the negative SI scores demonstrated a much greater range of scores compared to positive.

On dividing this subscale of children into their respective cohorts 10 belong to the epilepsy group and 10 to the non-epilepsy, Table 26. The values between the two groups were very similar and were closely linked to the overall means illustrated in the table above.

Table 26: SIP scores for separate cohorts at T0

Non epilepsy (Epilepsy)	Positive SI	Negative SI	SE
Mean	53.46 (52.89)	30.71 (29.92)	7.74 (8.99)
Median	55 (54)	31.00 (32.00)	4.00 (4.00)
Mode	47.00 (47.00)	14.00 (17.00)	5.00 (6.00)
Range	35 (30)	50.00 (54.00)	6.00 (6.00)

Administrative issues in obtaining further SIP questionnaires from the distributors occurred in a lack of copies for use at T6 data collections. Therefore no follow-up data can be presented for this questionnaire. This is further discussed in the discussion and limitations section of this chapter.

5.6.14 Paediatric Quality of Life Inventory Analysis

The PedsQL is a quality of life measure to be completed by children 6-16. Therefore, for this section of the results the sample size is n=26. This is due to the fact that not all children participants of this study fulfilled the age criteria for which this questionnaire is validated. Its total score ranges from 0-100 with higher scores indicating a greater quality of life.

The histogram below, Figure 63, illustrates the skew of the data collected from all the appropriate children. The range is spread from 40-100 with a mode of 70.

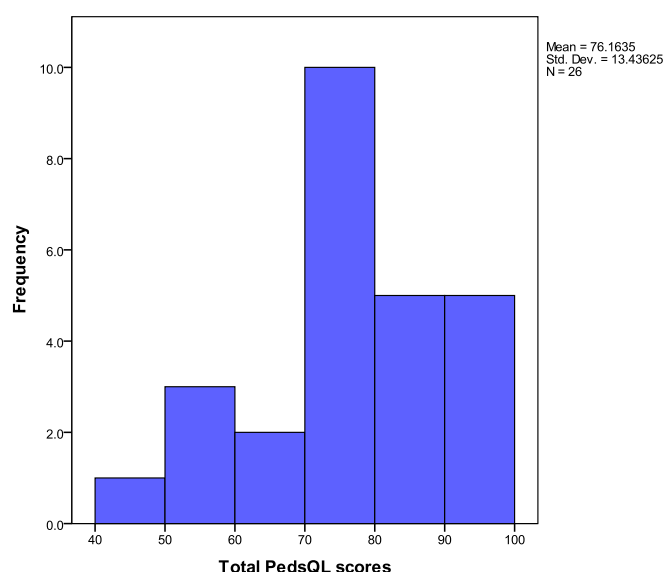


Figure 63: Histogram of total Peds QL scores from the total sample population at T0

Table 27: Descriptive statistics for total sample PedsQL data at T0 (n=21)

Descriptive Statistics					
		Paediatric quality of life physical functioning	paediatric quality of life emotional functions	paediatric quality of life social functioning	paediatric quality of life school functioning
Mean		83.301	68.654	84.231	68.462
Median		85.940	72.500	90.000	70.000
Mode		100.0	65.0 ^a	100.0	70.0
Range		62.5	90.0	55.0	65.0
Percentiles	25	74.225	53.750	73.750	55.000
	50	85.940	72.500	90.000	70.000
	75	96.881	86.250	100.000	80.000

a. Multiple modes exist. The smallest value is shown

Descriptive statistics were then conducted upon the component subscales, shown in table 25. This breakdown indicates that the emotional functions scale had a much greater range compared to the others. But it was also noticeable that the school and emotional functioning scales had a lower median and mean compared to the other subscales.

Of these 26 children, 11 belonged to the epilepsy cohort and 15 belonged to the non-epilepsy. Analysis of total scores was carried out based on cohort allocation and is shown in Figure 64.

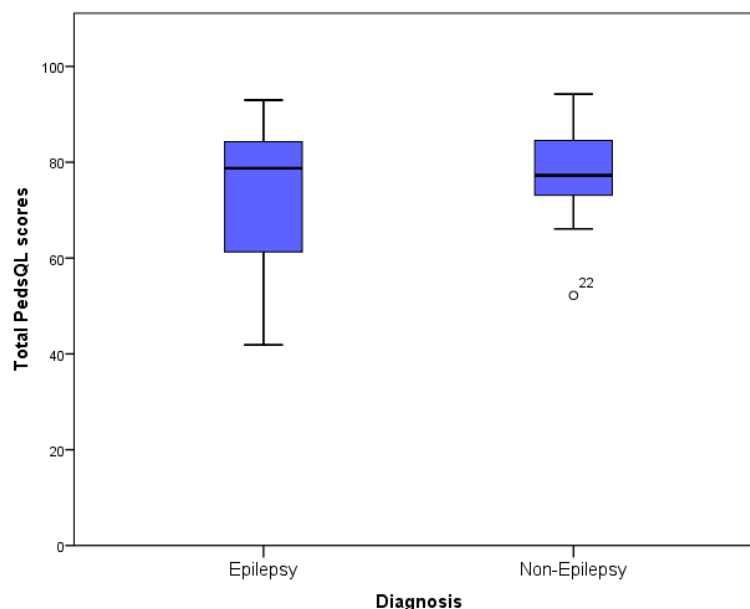


Figure 64: Box plot graph of total PedsQL scores between the cohorts at T0

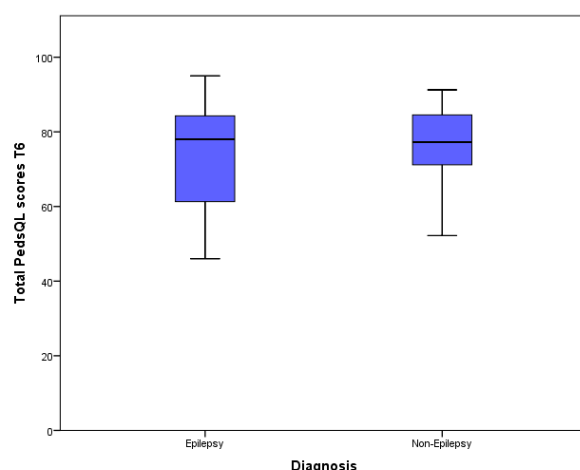


Figure 65: Box plot graph of total PedsQL scores between the cohorts at T6

In addition to graphing the differences of distribution of the cohorts, Figures 63-65, non-parametric statistical analysis was conducted to determine if there were any significant differences between the cohorts. Figures 66 and 67 below indicate that this was not the case for T0 or T6. This suggests that there was no significant difference in the quality of life scores of the children at time of presentation or at time of diagnosis.

Hypothesis Test Summary				
	Null Hypothesis	Test	Sig.	Decision
1	The medians of Total PedsQL scores are the same across categories of Epilepsy Status.	Independent-Samples Median Test	1.000 ^{1,2}	Retain the null hypothesis.
2	The distribution of Total PedsQL scores is the same across categories of Epilepsy Status.	Independent-Samples Mann-Whitney U Test	.775	Retain the null hypothesis.

Asymptotic significances are displayed. The significance level is .05.

¹ Exact significance is displayed for this test.

² Fisher Exact Sig.

Figure 66: Mann-Whitney U test results for cohort differences between total PedsQL scores at T0

Hypothesis Test Summary			
	Null Hypothesis	Test	Sig. Decision
1	The distribution of Paediatric quality of life physical functioning is the same across categories of Epilepsy Status.	Independent-Samples Mann-Whitney U Test	.610 ¹ Retain the null hypothesis.
2	The distribution of paediatric quality of life emotional functions is the same across categories of Epilepsy Status.	Independent-Samples Mann-Whitney U Test	.646 ¹ Retain the null hypothesis.
3	The distribution of paediatric quality of life social functioning is the same across categories of Epilepsy Status.	Independent-Samples Mann-Whitney U Test	.109 ¹ Retain the null hypothesis.
4	The distribution of paediatric quality of life school functioning is the same across categories of Epilepsy Status.	Independent-Samples Mann-Whitney U Test	.237 ¹ Retain the null hypothesis.
5	The distribution of Total PedsQL scores T6 is the same across categories of Epilepsy Status.	Independent-Samples Mann-Whitney U Test	.721 ¹ Retain the null hypothesis.

Asymptotic significances are displayed. The significance level is .05.

¹Exact significance is displayed for this test.

Figure 67: Mann-Whitney U test results for cohort differences between total PedsQL scores at T6

5.6.15 Stress Scores in Relation to other Covariates

Spearman rank correlation analyses were conducted to identify the strength of any correlation between carer stress and other stress covariates. These were calculated for all the variables of which there was a total or overall score usable to determine association. The correlation coefficients between the sets of variables are summarised below in Table 28.

The strongest correlations with stress, as measured by correlation to PIP stress scores were found between carer health, child behaviour and child HRQOL

Table 28: Summary table of spearman rank correlations between carer stress and carer covariates

		Correlation with Stress Measure	
Covariate	Covariate measure	PIP-f	PIP-d
Carer health	Total GHQ	0.50	0.46
Carer support	Total FSS	-0.18	-0.10
Carer needs	Total FNS	0.35	0.31
Carer LoC	Rotters LoC	0.08	0.08
Family functioning	FACES Total circumplex ratio	0.13	0.09
Family Communication	FACES family communication scale	0.02	-0.16

Child behaviour	Total SDQ	0.51	0.44
Child HRQOL	Total PedsQL	-0.50	-0.48

5.6.16 Stress Scores in Relation to Demographic and Epilepsy Data

Stress Scores and Demographics Correlation

Table 29: Summary of the correlation between carer stress scores and their demographic information

	Correlation with stress measure	
Demographic variable	PIP-f	PIP-d
Carer age	-0.24	-0.13
Carer gender	0.08	0.12
Carer education	0.17	0.16
Family structure	0.09	0.06
Number of children	-0.14	-0.12
Age of affected child	0.19	0.09

Table 29 above indicates the degree of correlation between the stress scores and demographic variables. As shown, none of these variables demonstrated a strong correlation, although a weak negative association was found between carer age and their reported stress.

Table 30: Summary of the correlation between carer stress scores and their child's 'fit' information

	Correlation with stress measure	
Epilepsy variable	PIP-f	PIP-d
Frequency of event	0.59	0.54
Time elapsed since first event	0.20	0.17

Table 30 compares the data from the suspected seizure characteristics and reported stress scores. As might be expected the frequency of these events had a stronger positive correlation whereas time from first event to presentation had only a weak positive association.

5.6.17 Stress scores from different measures

The key variable: perceived stress, was measured using 2 tools. These were the PIP and SSSQ. A simple scatter plot, Figure 68, followed by regression analyses were conducted to determine the correlation between the scores of the 2 measures

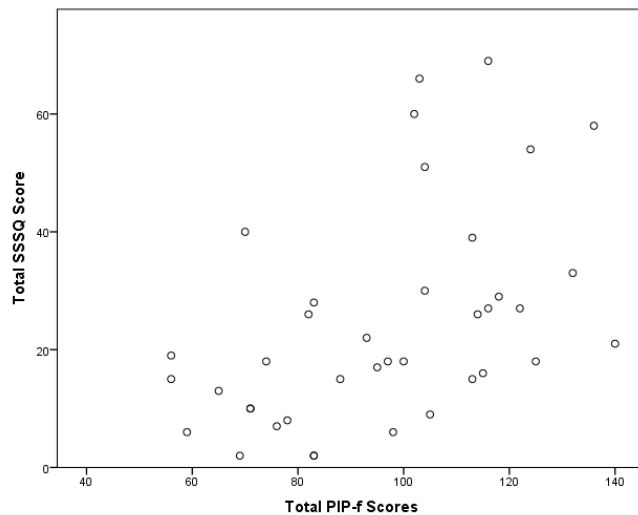


Figure 68: Scatter plot graph of total PIP frequency scores and SSSQ total scores from T0

The SSSQ is a novel measure, developed and used for the first time in this research. In order to gain an idea of its potential for further use its scores were compared to that of the well validated PIP measure. The correlation coefficient between these tools was found as $r=0.38$. This association may also be seen on the graph of Figure 68.

5.7 Discussion:

5.7.1 Recruitment and Follow-up Discussion

The overall rate of recruitment for this study was 45% and of the 196 potential participants approached, via postal information pack about the study, 108 of them did not consent for this study. As illustrated in Table 15, the most frequent reason potential participants were not included into the study was due to non-attendance at the scheduled appointment, due to either cancellation or they simply did not attend. It seems surprising that 36% of carers did not bring their child to clinic particularly with a potential diagnosis of epilepsy. Attempts were made by the researchers to recruit these families at their rescheduled clinic dates but this was not always possible. Although it may be the case that the 'suspicious' events resolved and therefore they did not attend it may however suggest that there are some children in the community experiencing epilepsy without being given the diagnosis or offered the appropriate treatment.

It is also apparent that a quarter of potential participants were not included in the study because they did not consent. This is discussed separately in the limitations section of the discussion. It is worth pointing out that anecdotally, having conducted many recruitments over the course of this year some of the reasons given to me by parents for not wanting to participate were 'because I'm too stressed to sit down and take part'. This is relevant as reasons such as this may be a source of bias to the results of this study. It is however difficult to reduce the impact of this bias because parents must make their own decisions regarding study participation and researchers should not persuade or coerce people into the study to maintain ethical guidelines as set out by the GMC.

The time between the first and second data collection points was variable. This was mainly due to fitting the follow-ups around the scheduling of the patient's return clinic appointment or when they would next be visiting AHCH. It was organised like this in order to minimise the burden on the participating families. As mentioned in the methodology, questionnaire packs were sent via post to a family's home address if they had no scheduled return clinic appointment or if they had not attended their follow-up appointment. Therefore in some cases the length of time between data collection points was limited by the time taken by carers to return the questionnaire packs.

Although the mean period between data collection points was 6.4 months it should be acknowledged that the wide variability may impact the results. All follow-ups were conducted after 3 month from the first data collection which minimises any potential practice effect. Differing periods of time between completing the first and second batches of questionnaires however, may affect the stress levels and other covariates being recorded. This may be

because carers will have had more/less time to come to terms with the information and potential diagnoses given at clinic and therefore had different amounts of time with which to adapt or develop coping responses to these potential stressors. Habituation may also be a factor. Therefore it is relevant to consider this range of follow-up period as a potential factor causing bias to the results.

As illustrated in Table 16 not all families completed both questionnaire data collection time points. Of the 45 who only completed the first appointment 17 were lost to follow up. This may be understandable given the prospective nature of the study and the time gap between appointments. 28 carers however were not followed up. This was unfortunately due to AHCH being unable to fund another M.Phil student to continue this project after my 12 months finished. Attempts were made to account for a research nurse to continue but this was sadly not possible.

5.7.2 Demographics Data Discussion

Of the 39 families who fully completed the questionnaire battery for both time points 82% of the primary caregivers were mothers. Whilst the average age of all the carers was reported as 34.9 years. 93% of the study participants identified themselves as Caucasians. The majority of caregivers' education status was completion of college. Although it was expected that mothers would form the majority of primary carers, it was interesting to note that 15% were fathers. This is a higher percentage than recorded in prior studies investigating stress in parents (Hua-Huei and Liang-Po, 2008). It was additionally surprising that only 7% of the sample did not report their ethnicity as Caucasian, given the multicultural nature of Liverpool and some of the surrounding areas.

Several factors pertaining to the child were also collected. Of the 39 children, the gender ratio of both the overall sample population and the ratios between the cohorts were broadly similar. Additionally, the mean ages of participating children between the cohorts was found to be 7.4 and 7.7 years. A further point of particular note regarding child age is that out of the 39 families involved in the study only 26 of the children satisfied the age criteria for completion of child oriented questionnaire. This significantly impacts the sample size for these 2 questionnaires, the PedsQL and the SIP-C. Therefore this may also affect the reliability of the results recorded by these two instruments.

Statistical analysis of the demographic data between the cohorts was predominantly done using Fishers exact test. This method was chosen because it is appropriate for use when the collected data is categorical and the cell count is less than 5 and in this case both of these criteria apply. For those categories where the cell counts were all greater than 5 a chi square test was used as it is more appropriate for larger numbers. The results of this analysis indicate that there was no statistically significant difference between the epilepsy and non-

epilepsy cohorts. This implies that it may be valid to make comparisons between the Confirmed Epilepsy and Non-Epilepsy cohorts.

5.7.3 Discussion of 'Fit' Questionnaire results

Using the 'Fit' questionnaire, parents reported the estimated frequency of their child's events. The mode for cohorts (which were determined retrospectively *i.e.* not known when parents completed this survey) was the 1-2 month category for the epilepsy group compared to the 1-4 per month category for the non-epilepsy. The medians for both cohorts lay were 1-2 per month. However 3 carers (7%) reported that these events were occurring greater than 3 times a week.

Data collected regarding the period of time since first onset to presentation at neurology outpatient clinic was extremely variable. Overall, most carers reported a period of 3-4 months although it is interesting that 50% of the carers who were later assigned to the epilepsy cohort presented between 1-2 months compared to just 7% of the non-epilepsy carers. It is also of note that 21% of carers said that there was a greater than 6 month period between their child's first queried seizure and attendance at clinic. This was a particularly concerning finding and therefore using the recruitment scheduling database for this study a rapid analysis was made between date of referral and date of scheduled clinic appointment. This revealed that the mean waiting time between referral and outpatient appointment was 14.9 days with a range between 11 and 23 days (DNA rescheduled appointments not included). Whilst this is reassuring with regard to the Alder Hey scheduling system, it still does not explain these findings. Although a potential reason for this may be due to the self-report nature of this item and the participants recall of events.

Three carers reported that their child had some form of investigation before their first attendance at outpatient clinic. It was considered whether or not to exclude these carers however following discussions with supervisor and lead investigator Andrew Curran it was decided to keep them in the sample as they still at presentation had no diagnosis for their child's events. It should be noted that the experience of prior investigations for these potential seizure events may have impacted carer stress scores and therefore add a small element of bias to these results.

Questionnaire completion appointments were organised around the participant families in order to maximise recruitment. The aim was for these appointments to occur prior to the child's clinician review. This was not always possible, approximately 20% of all appointments occurred after seeing the medical team. Key reasons for this were family time commitments, researcher availability, clinic scheduling and room availability. For 27% of all appointments the timing is unclear. The principal reason for this was to the 2 previous researchers collecting

data did not record this information. These issues have been recognised as important factors potentially affecting the data and are therefore further discussed in the limitations section of this thesis.

5.7.4 Discussion of Paediatric Inventory for Parents results

Firstly, from examination of the histograms presented by Figures 14 and 15 in the results section it is apparent that the data does not exhibit a Gaussian distribution. This suggests that there is an element of skew in the data. This was to be expected due to the nature of the study population. Recruitment of caregivers potentially facing their child's new diagnosis of epilepsy may be assumed to experience greater levels of stress than a representative sample of the general population. However, this finding may also reflect the small sample size of this study.

Secondly, the histograms also suggest that at T0, 59% and 56% of caregivers were experiencing high levels of stress based upon PIP-f and PIP-d domain scores. This is determined by the standard cut-off value of 90 which is illustrated on both graphs by the bold orange line situated on the x-axis. The score of 90 has been used in concordance with work by Streisand, one of the creators of the PIP.

The differing ranges shown between the PIP-f and PIP-d scores are also worth note. As illustrated again by figures 13 and 14, overall caregiver had a much narrower range of scores on the frequency domain, whereas the range for the difficulty domain was greater. Ranges were 84 and 98 for PIP-f and PIP-d respectively. This information may suggest that it is not the frequency of events that prove stressful for the caregivers but more the difficulties experienced as a result of them.

Correlation analyses between total PIP-f and PIP-d domain scores were carried out at both T0 and T6, illustrated graphically in Figures 17 and 18. Linear regression scores indicated a correlation of $r = 0.65$ between the domains at T0 and a correlation of $r = 0.75$ at T6. Although strong positive correlations such as these were expected, the explanation for the significantly increased correlation at T6 is not known.

In addition to examining the data of the sample overall, comparative analysis was also carried out between the epilepsy and non-epilepsy cohort. Graphical representations of total PIP-f and PIP-d scores between the two cohorts were conducted both at T0 and at T6. The previously discussed cut off score of 90 was applied for clarity. Following this Mann-Whitney U tests were carried to identify any significant differences between the cohorts. This non-parametric method of analysis was chosen to account for the potential skew of the data set

and to reflect the categorical nature of the epilepsy variable. The results of these, as shown in Figures 16 and 18, indicate that at T0 there is no statistically significant difference between PIP scores of the epilepsy or non-epilepsy cohort. However, at T6 there is a statistically significant difference between the PIP scores of the two cohorts ($p < 0.05$).

These findings suggest that carer stress scores at time of presentation, *i.e.* prior to diagnosis, are equivalent and that following their child being given a diagnosis of paediatric epilepsy caregiver stress levels are significantly increased compared to those carers whose children do not go on to have epilepsy.

Intuitively this is what was expected to be found, given the wealth of literature documenting increased stress levels in carers of children with epilepsy. However, given the relatively novel study design it is difficult to directly compare these results with existing research. Principally because prior studies researching stress in epilepsy have used control groups of either healthy children or children with different diseases that may not have any similar symptomology or course to epilepsy. Interestingly however the mean and median values constructed from this data are greater than those found in other studies using the PIP as an outcome variable, see table 4. This may be, in part, down to differences in the paediatric condition being investigated such as sickle cell anaemia, diabetes and bladder extrophy. However, in a 2010 study by Wagner involving children with epilepsy, the means for the PIP domain scales were 86.1 and 77.6 (Wagner et al.) for the PIP-f and the PIP-d compared to 96.13 and 92.62 found with this study. It is worth noting that the sample size was $n=9$ and this small study population could introduce a degree of skew which may explain the differences in PIP scores.

5.7.5 Discussion of Semi-Structured Stress Questionnaire Results

From the histograms of the SSSQ carried out on the total sample, it is again made clear that there is an element of skew in the dataset for this sample population. This is in concordance with the previous findings from analysis of the PIP. These histograms also show that the range of scores recorded by the carers is also very wide. This may reflect the diverse nature of the families included in this study, as per the family resiliency model's acknowledgement of the individual characteristic that determine how families may react to stress.

The reference lines on the x axes of Figures 24 and 25 indicate the cut-off score of 10 which is used to determine the carers experiencing greater levels of stress. The T0 histogram suggests that at time of first presentation 79% of caregivers had high levels of stress and 76% were experiencing high levels of stress at T6. With regard to the SSSQ as a novel tool with a lack of published data detailing its psychometric and diagnostic properties it may be warranted to investigate the decision of using a cut-off score of 10 to indicate significantly

high stress levels. Raising this to a score of 25 or even 30 may have a positive impact on the specificity of this tool. This is inferred based upon increasing the SSSQ cut-off to 25 meaning that the percentage of carers identified with increased stress levels would be comparable between the validated PIP measure and the novel SSSQ.

Box plot graphs were carried out for the separate cohorts to examine the spread between the epilepsy and non-epilepsy cohorts. Figure 24 indicates that carers in the epilepsy cohort had a wider range than that of the non-epilepsy cohort.

Following the box plot analysis a Mann-Whitney U test was also conducted as a non-parametric method to examine the differences between the median and distributions between the 2 cohorts. The outliers present in the relevant box plot graph were left in. It is felt to be an appropriate action given that the Mann-Whitney U test functions by assigning a rank to each data value meaning that any outliers are unlikely to affect the validity of the findings. The results of this analysis showed that there was no statistical difference between the cohorts at T0. This correlates with the results obtained with the PIP, further reinforcing the assertion that stress levels at time of presentation are comparable between the carers of the children that do and do not go on to receive a diagnosis of epilepsy.

Statistical testing of the T6 data however, shown in Figure 29, found that differences between the 2 cohorts at this time were statistically significant $p=0.048$. This indicates that stress levels are higher in carers of children with epilepsy than those of children who do not. This result was expected and is in agreement with previous work investigating stress in carers of epilepsy. By having a control group who also went through a similar experience i.e. their child having queried seizures, the associated uncertainty and referral to a large tertiary paediatric centre it may suggest that is predominantly the diagnosis and its associated sequelae that are the main stressors for these carers rather than the actual event/seizures themselves.

Despite the findings of the SSSQ data to be broadly what we anticipated it is difficult to correlate these with the prior literature. This is primarily because it is a novel tool, first used in this study. It is however, encouraging to note its positive correlation with the widely validated PIP measure, as shown in Figure 68.

5.7.6 Discussion of General Health Questionnaire-28 Results

Analysis of total GHQ scores found that 36% of responding carers were scoring within the range for psychological distress. This implies that in terms of their mental state, approximately one third of carers may be experiencing some symptoms of psychiatric illness. This is significantly lower than some other studies involving carers of children with epilepsy such as (Sreeja et al., 2009) and Behrouzian (F and S, 2010) which reported 55% and 67%

respectively. These differences may be down to several factors, all used relatively small sample sizes $n = 39, 35$ and 50 . Additionally geographical and cultural confounders may have had an effect due to the different populations upon which these studies were carried out.

Of the 4 subscales which form the total score, it is apparent that the 'somatic symptoms' and the 'anxiety and insomnia' scales were rated more frequently as they had higher mean and median values whilst the 'depression' subscale was the least scoring as the mean, median and mode scores were all zero.

It was interesting to note that conducting the same graphical analysis on the T6 data, the median total GHQ score of the epilepsy group remained the same, while that of the non-epilepsy dropped. It is suggested that this may be as a result of the lesser stress that these carers are experiencing because based on the PIP and SSSQ analyses we have already determined that carer stress was significantly lower for the carers of children without epilepsy than those with at T6.

It should not be overlooked that even at T6, 21% of carers still had scores greater than 5 suggesting that just over one fifth of parents are still exhibiting signs of mental ill health. Information as to whether or not carers were receiving any kind of medical attention regarding these symptoms was not collected by this study but it would be of interest, as it may be considered logical that optimising the caregivers health may have a knock-on effect on decreasing their stress levels and improving the wellbeing of themselves and their families.

5.7.7 Discussion of Rotter's Locus of Control results

From the results described in section 5.6.7 it is seen that distribution of Rotters LoC scores for the total sample of carers was wide- ranging from 7-20. As the maximum potential score is 23 it suggests that there are more caregivers with highly external loci than there are with highly internal loci. From this histogram a median score of 12 was determined and therefore based on the literature existing, this was used as the cut-off value for determining a carer's LoC.

Box plot graphs of the T0 data indicated that both cohorts exhibited a similar range of scores. It was interesting to note however that the epilepsy cohort exhibited a narrower inter-quartile range than that of the non-epilepsy- spanning 10-14 compared to 9-16. Furthermore the medians for cohorts were 13 for the epilepsy group compared to 11 for the non-epilepsy i.e. each lying on the other side of the cut-off value of 12. These differences however, were not found to be significant.

When considering the data collected from T6 it was apparent that the overall spread of data was the same as that at T0 however it was slightly skewed downward, spanning from 6-19.

Interestingly the median values for both groups were equal to the overall median of the total sample and fell onto the same value of the cut-off boundary. This suggests that at T6 not only were the two cohorts comparable in terms of the LoC scores, but that regardless of whether their child went on to have epilepsy carers reported mainly balanced and healthy locus of controls.

These findings appear to differ to some existing work, however none was found that were exclusive to parents of children with epilepsy. Work by (Perrin and Shapiro, 1985) reported that parents of children with chronic medical disorders reported higher external and lower internal loci of control. Their work also demonstrated some similarities with this research namely their median value was also 12 and their results were closely grouped around the centre of the spectrum with fewer respondents scoring at either extreme.

Although these findings are indeed encouraging, they were not necessarily expected. It was previously queried by this author as to whether LoC scores for the epilepsy cohort at T6 would be significantly higher than their non epilepsy counterparts based upon the unpredictable nature of the disease and the uncertainties previously recorded in studies investigating parental stress and epilepsy. Despite the unexpected nature of the results it is useful and positive information because there is no clear method by which it is possible to change an individual's LoC. Therefore if scores had been shown to be significantly different between cohorts it would be difficult to organise a helpful intervention by which to alter the carers loci of control.

5.7.8 Discussion of the Brief COPE results

Analysis of the Brief COPE T0 did not reveal any significant finding, although there were some possible trends which may indicate areas for future work with larger sample sizes. Having viewed these potential differences on the graphs, represented by Figures 37-39, Mann-Whitney U tests were conducted to determine their significance. However, as previously mentioned, they confirmed that there was no statistically significant difference in coping scores between the cohorts at T0. Although it was expected that no differences would be found between the cohorts for any of the covariate measures it is still relevant to note that at time of presentation carers regardless of whether or not their child went on to have epilepsy were utilising comparable coping strategies.

Breakdown of the descriptive statistics for the individual subscales formed by the brief COPE suggests that 'self-distraction', 'instrumental support' and 'acceptance' scales were the most highly scoring based upon their mean and median values illustrated by Table 22. This indicates that they are the most frequently used coping strategies employed by these carers. Interestingly, each of these subscales belongs to a different overall coping strategy.

Identical procedure was then carried out for the T6 brief COPE data. These analyses revealed that there was no significance in the different medians and distributions between the cohorts for their problem focused and emotion focused scores. However at T6 it was found that the epilepsy group reported significantly higher dysfunctional coping scores. A potential reason for this may be that having received a new stressor, a diagnosis of paediatric epilepsy, the carers have not yet had sufficient time with which to adapt and develop appropriate coping strategies whilst for the non-epilepsy cohort their situation has not changed and therefore their coping methods remain predominantly functional

Although the findings from the Brief COPE Inventory were not found to be significant it is encouraging to see that the results had some parallels with existing research. Work by (Cooper et al., 2008) indicated that carers, used problem focused coping measures foremost, followed by emotional and then dysfunctional coping strategies. This work had a sample size of n=125 but a similar trend was visible in our much smaller values.

5.7.9 Discussion of Family Support Scale Results

As shown in the FSS results section, the spread of total FSS scores at T0 was broad ranging from 20-62 out of a maximum of 90. For the T6 data this range was marginally smaller from 20-55. This may reflect a variety of things. Firstly, that there is heterogeneity in the degree of support sought out overall among the primary carers. Or secondly, that there may be heterogeneity in the sources of support available to different families. It may also likely be a combination of the two.

On separation of total FSS scores by epilepsy status at T0, it is apparent that the ranges between the cohorts were broadly similar. The medians for the cohorts were 35 and 32 respectively for the epilepsy and non-epilepsy groups. Both these and the distributions between the cohorts at T0 were found to be not statistically different. This was expected and appears logical given at T0 neither cohort had a diagnosis.

However, differences between the cohort's total FSS scores at T6 were also found to be insignificant. This is not what I had necessarily expected to find. Given that stress scores were shown to be significantly increased in the epilepsy cohort it seemed logical that this group of carers would be likely to reach out for greater levels of support than those confirmed to not have epilepsy. From this data it appears that this was not the case.

Analysis of the 5 subscales was also carried out at T0. Results of these indicated that the levels of informal support received by the carer were the most variable of the available supports. Additionally it exhibited the greatest mean score which suggests that they were

considered most helpful by the carers. Unfortunately however, on segregation of the data into 5 scales the numbers were too small to perform any meaningful statistical analysis without reading too much into the data. This domain should still be of interest for future research with a larger cohort because family support is one of the few areas in which healthcare staff may have the opportunity to impact the stress experienced by carers.

These observations are similar to those made in prior work. Work by (Levy, 1996) reported a wide range of responses in the sample population of n=132 mothers of children with 'mental retardation'. This suggests that the heterogeneity of our responses may not be an unusual finding. Levy's research however, also showed that carers scored professional support most highly. This is of interest as it is an area in which healthcare staff may be able to decrease carer stress.

5.7.10 Discussion of Family Needs Survey Results

The total FNS scores as reported by the carers ranged between 0 and 17. Although the overall mode for the population was 0 within this sample 82% of carers expressed some level of need at T0. This is expected and understandable given that at time of presentation all carers are in a similar unknown situation. This is further shown as the most frequently reported needs were those pertaining to information and resources.

Counselling needs were the most infrequently reported, at T0- 8 and at T6 only 5 carers indicated a definite need in this area. This suggests that when considering future interventions targeted at this type of population they may be of greater use to a wider population if they were centred on providing better information to carers. That is not to undervalue the needs of those parents with counselling concerns however. In fact, all parents when carrying out the questionnaires were informed of the psychology and counselling supports available to parents and carers at Alder Hey should they require it.

The lack of significant difference between FNS scores between the T0 and T6 data does not strictly correlate with other works using FNS at multiple data collection points. In fact, (Trute and Hiebert-Murphy, 2005) reported a p-value of 0.01 for the difference in FNS scores between T1 and T2 occurring 6 and 18 months after entering child disability services. Trute's study may not be directly comparable as it collected data on general childhood disabilities rather than a single condition such as epilepsy. However this enabled greater sample size, n=111, and increased the likelihood of finding significant results. At time of writing it was not possible to find previous research using the FNS in the context of paediatric epilepsy alone.

5.7.11 Discussion of FACES IV results

Analyses of the FACES total circumplex ratio data, a measure of family functioning, revealed that both cohorts exhibited similar medians and ranges at both T0 and T6. All carer scores were ≥ 1 which indicates predominantly healthy levels of family functioning which in terms of the overall family's wellbeing is distinctly reassuring. Although the inter-quartile range at T0 was broader in the epilepsy cohort suggesting that there is greater heterogeneity of family functioning within this group statistical tests indicate that there was no significant difference between the carers at time of presentation. This is in concordance with the results of the PIP and SSSQ which also showed no differences in the cohorts at this time. This also correlates with family stress models as the families have yet to determine the nature of their child's events and therefore have not yet had to adapt or alter their role functions within the group.

For the T6 data, there was still a broad overlap between the distributions of the 2 cohorts' total circumplex ratio scores. The median values were lower than those at T0 but still fell closely (1.93 compared to 2.06 for the epilepsy and non-epilepsy groups respectively.) These observations were confirmed by Mann-Whitney U testing determining that there was no statistically significant difference between the total ratio scores of these cohorts. This finding was unexpected given the existing literature purporting family functioning to be a mediating factor of family stress. However, the sample size of this study may have affected the strength of any potential trends to be observed.

The family communications scale is formed from 10 items within the FACES tool and the raw scores were translated into percentage scores. This is in concordance with existing FACES literature and allows for easier interpretation of results. Ranges at T0 were similar between the 2 cohorts: 10-97% and 10-99% for the epilepsy and non-epilepsy groups respectively. The inter-quartile range for the epilepsy group was broader than that of the non-epilepsy and also had a lower median. However, Mann-Whitney U testing indicated that there was no statistically significant difference in family communication between the 2 cohorts on this scale. This is logical, given the assumption that all carers are in a similar situation *i.e.* all at first presentation for queried seizures in their child.

Graphs of the T6 family communications percentages also broadly showed the same distributions between the 2 cohorts, despite the presence of one outlier in the non-epilepsy cohort. The medians for each group were also slightly lower, 68% and 74% for the epilepsy and non-epilepsy groups respectively. Although these differences were observed, statistical analyses showed that in fact these were no significant. This implies that following diagnosis there was no significant difference in the family communication of those which have a child with epilepsy and those that do not. Similarity between the 2 cohorts on this scale may partly be due to the timing of the second data collection point. Carers will have only just received the

new diagnosis but it has not yet been integrated into the daily lives of the family and the level and quality of the family member's communication.

Data from the family satisfaction scale has been presented in a similar manner to that of the family communications scale based on the same reasoning. This scale also indicated that in both cohorts primary carers exhibited a wide range of family satisfaction levels at T0. The median for the epilepsy cohort was again lower than that of the non-epilepsy but as evidenced by the Mann-Whitney U analysis neither this nor the smaller inter-quartile range of this cohort was significantly different to that of the non-epilepsy cohort.

Potential reasons for satisfaction score observations are again in keeping with those for the communication scale, as the families, regardless of what their child's diagnosis turns out to be, are all having a similar experience at their initial presentation. It is suggested that at time of diagnosis, although a significant difference was expected, it may be down to the minimal time period between receiving the diagnosis and completing the questionnaires that not enough time had passed for the diagnosis of epilepsy to affect the carers satisfaction scores. The wide range shown in the overall sample at both time points may be a reflection of the different levels of family resiliency amongst other characteristics which are highlighted in the models of family stress discussed in Chapter 1.

Whilst this research did not find any significant difference in family factors between the epilepsy cohort and the non epilepsy controls previous work has demonstrated this finding. A review by (Rodenburg et al., 2005) highlighted that families with children diagnosed with epilepsy had significantly lower levels of communication and esteem when compared to families of children with asthma, $p=0.01$.

5.7.12 Discussion of Strengths and Difficulties Questionnaire results

The SDQ was used in this study for a proxy measure of child behaviour. Prior to the carer completing this questionnaire they were advised by the attending study researcher to think only of the child referred to neurology when scoring rather than any other children that they may have. Analysis of the total SDQ scores for all 39 carers at T0 indicated that there is a wide range behaviour exhibited by these children. Reference lines on the histogram indicate that 72% of carers reported scores that are interpreted as abnormal or difficult.

This large proportion of carers reporting their children as having difficult behaviour is surprising. The reference ranges and values were determined by testing on 10298 parents in the UK (Mind, 2011). The mean given for the normal population was 8.4 which is in stark contrast to the overall means of 22.2 and 21.3 at T0 and T6 of this research respectively. A variety of factors may have impacted this difference for example the small sample size of this

thesis may have given an artificially high result. Sample size issues are further discussed in the limitations section. The study population of Liverpool parents may have also affected the scores and their tolerance of what constitutes poor behaviour may vary. This 72% does however correlate with the findings of Austin, whose work suggested that children with chronic disease exhibit increased levels of difficult behaviour which is linked to parental stress.

When these total SDQ scores were broken down into cohorts it is visible that the inter-quartile ranges for both cohorts lie above the reference lines indicating borderline and abnormal behaviour. This is interesting because there is much previous literature supporting the notion that there is a correlation between epilepsy and behavioural problems (Austin and Caplan, 2007). It has further been the topic of recent debate as to whether the behavioural problems precede the diagnosis of epilepsy (Austin et al., 2001). Therefore, it may have been expected for the epilepsy group to a higher level of behavioural problems but in fact no statistical significance was identified between the cohorts. This may, in part, be down to what individual parents and carers consider to be difficult behaviour. Additionally it may reflect the nature of the SDQ and a potentially high sensitivity. Further work such as a meta-analysis of previous studies using this tool could be evaluated with regard to the cut-off values used for determining 'borderline' and 'abnormal' behaviour.

At T6 the ranges observed for the total SDQ scores were calculated as 10-36 and 11-33 for the epilepsy and non epilepsy cohorts. Despite both of these groups illustrating overlapping inter-quartile ranges and similar median values- 23 compared to 21, the breadth of the epilepsy group was marginally larger indicating a greater range of childhood behaviour in the children diagnosed with epilepsy. Again however, statistical testing revealed these findings to be insignificant. It is hope that with ongoing follow ups and a greater sample size in the future smaller effect sizes will be needed to determine significance.

5.7.13 Discussion of Self Image Profile Results

Results from the SIP-C and SIP-A were compared as suggested by (Butler, 2001), hence the scorings for both versions of the questionnaires are equivalent. For the total sample (n=20) at T0 the mean and median scores for the positive SI scale were greater than that of the negative. This indicates that overall, at time of presentation, the children viewed themselves as having more positive attributes than negative. The range of negative SI scores however, was larger than that of the positive, 53 compared to 31, implying that there was a greater range of negative attributes that children perceived in themselves compared to positive ones.

However, based upon the low self difference scores it is apparent that broadly speaking the children completing this questionnaire had good self esteem. This is evidenced by a mean SE

of 8.92 indicating that there was only a small difference between the way that children viewed themselves and the way that they wanted to be.

Upon dividing the SIP scores based on the child's epilepsy status further descriptive statistics were conducted as seen in Table 26. These illustrated the same broad observations as visible in the overall data table however it was of note that the epilepsy group reported a lower positive SI median (54 compared to 55) and a greater negative SI median (32 compared to 31) than that of the non-epilepsy cohort. Although these differences were not found to be significant on Mann-Whitney U analysis, it is interesting because at T0 *i.e.* time of presentation no diagnosis for these potential seizure events had been ascertained. Therefore theoretically there should be little reason for the children to have different self-images. Also the measure of self esteem found that generally speaking all the children's scores indicated a good self esteem, the epilepsy group had a larger mean (8.99) than that of the non-epilepsy (7.74). This suggests that the epilepsy group may have a lower self esteem as they showed a larger gap between how children see themselves and how they want to be. The difference between these scores was not found to be significant; however, this would be an interesting finding to bear in mind for future larger studies comparing these cohorts.

As may have been noted in the results section no T6 data has been presented for the SIP questionnaires. This was due to a delay in receiving a second batch of questionnaires from the suppliers. Therefore there is unfortunately insufficient T6 SIP data in order to report. This lack of data limits our understanding of any potential changes in the children's potential changes in their self image following their new diagnosis of epilepsy. This was expected to be an interesting finding of itself and also in how it correlates with the child's perceived quality of life scores. As a result, this was an unavoidable but acknowledged weakness of this work. By recognising it and its subsequent effect, it is hoped that this issue will not be repeated.

5.7.14 Discussion of Paediatric Quality of Life Inventory Results

The histogram of total PedsQL scores indicates that there was a range of scores across all the children between 41 and 96. Although the most frequent total quality of life scores were between 70-80, it is still of note that 15% (n=4) of children were rating their quality of life below 60. On further examination of the breakdown of these children's scores it appeared that their emotional and school functioning scores were lower than their physical and social functioning scores. This may suggest that interactions and comparisons between other children, as in a school environment, may be a factor in how children rate their own quality of life.

On segregating the total PedsQL scores based on the epilepsy outcome of the respective child, it was interesting to note the broader overall and inter-quartile ranges of the epilepsy cohort in comparison to those of the non-epilepsy at T0. However on statistical testing using Mann-Whitney U analysis, it was revealed that these differences were not statistically significant. This is unsurprising as the children have yet to attend a neurology clinic appointment. The lack of difference between HRQOL scores may be because the children of either group do not know the origin of these events that they have been having. It's postulated that the children, dependant on their age, may not even recognise at this point that such events are not part of normality.

The T6 comparative graphs between the cohorts again found that the epilepsy group exhibited the wider range and inter-quartile range. Importantly, as at T0 some children reported that their perceived HRQOL was as low as 45/100. Median values however were greater with scores of 78 and 77 for the epilepsy and non-epilepsy groups respectively. These differences were shown no statistical analysis not to be significant. However, it was interesting to note that overall the epilepsy group had a greater total HRQOL score than the non-epilepsy cohort, given the existing literature to the contrary. Explanations for this may be, as previously mentioned, due to the completion of questionnaires occurring very shortly after having received the diagnosis. The children who have been diagnosed with epilepsy may not yet have experienced how the diagnosis and its sequelae affect (or do not affect) their perceived quality of life.

Although no significant difference was found between the epilepsy and non-epilepsy cohorts regarding child quality of life previous research this is contrary to some existing work. (Devinsky et al., 1999) reported decreased child quality of life in adolescents diagnosed with epilepsy. This difference in results may be partly due to the participants. Adolescents may be more self-aware and have a greater knowledge regarding a diagnosis of epilepsy and its potential consequences, whilst younger children would not. The mean age of children in this study was 7.6 years, compared to a mean of 14.2 years in Devinsky's study.

5.7.15 Discussion of Stress-Covariate Analyses

For each of these correlation analyses a Spearman's rank test was used to calculate the correlation coefficient. This method was chosen based upon personal communications with Dr Lane. It is the most appropriate test to use because it makes no assumption of normal distribution of the data unlike other techniques. The PIP was used as the stress measure for these correlations because it is a previously validated tool. From examination Table 28 it is apparent that the total PIP-f domain consistently showed marginally higher degrees of correlation with the other covariates. Although these are not significantly different it may imply that the PIP-f domain is a slightly more sensitive scale.

Data collected from this study indicates that there is a correlation between carer stress and carer health. The correlation coefficients between the GHQ and PIP scales show a moderate degree of correlation between these 2 variables. This association suggests that there may be some value for using the GHQ as a quick screening tool in similar populations of parents. This could be a line of future investigation when attempting to identify 'at-risk' parents for interventional purposes.

Only a very weak negative correlation was identified between carer stress scores and levels of carer support. Although a lack of significant findings in the main analysis of the FSS results did precede this it was still an unexpected result. The negative correlation between carer support and carer stress is logical and consistent with existing theories it was expected that the association between these 2 variables would be much stronger. Unfortunately there is no obvious explanation for this although the small sample size may be a factor in this result.

Carer needs showed a weak association with carer stress scores. This is illustrated by the r values determined by Spearman's rank correlations. This correlation was a logical finding, however, as with the results of the FSS correlations it was perhaps expected that this correlation would also be stronger.

Analysis of the association between carer stress and their locus of control show negligible correlation between these 2 variables. This is highlighted by the relevant r values of 0.08 for both the PIP-f and PIP-d domains, which suggests that general locus of control is not strongly associated with carer stress levels.

Spearman's rank testing revealed a moderate strength correlation between carer stress and child behaviour scores this illustrated by the reported correlation coefficients of $r=0.51$ and 0.44 . The other moderate association between carer stress and child factors was the moderate negative correlation found between stress and child HRQOL, this is evidenced by coefficients of $r= -0.50$ and -0.48 with total PIP scores and the PedsQL. This finding is in keeping with prior research with found similar correlations, $r=0.50$ between proxy scored child QOL and parental anxiety measures. This association is also of interest due to existing literature evidencing the inconsistencies between child quality of life scores and the carers proxy scores of their child's quality of life in an epilepsy sample (Baca et al., 2010).

Overall carer health, child behaviour and child HRQOL covariates were found to have the strongest correlations with carer stress, suggesting that child variables may have greater associations with carer stress than variables pertaining to the carer themselves. This appears logical given the widely perceived belief that parents and carers put their children before themselves.

5.7.16 Discussion of demographic and epilepsy correlations and stress

As illustrated by Table 29 it is apparent that there were no strong correlations between any of the carer demographic variables and their reported stress scores. Carer age showed a weak negative association with stress, demonstrated by correlation coefficients of -0.24 and -0.13 for the PIP-f and PIP-d measures respectively. This seems logical as with increased age it may be that these carers have greater knowledge and parenting experience and are therefore less stressed in their role. It could, however, also be argued that the older carers may be those with older children and that the age of the child may be the explanation for the slight decrease in stress. This is not however borne out by the analyses for which no correlation was found between child age and carer stress scores.

Information about the children's suspected epilepsy or seizure events were also correlated against carer stress scores. As was reasonably assumed, frequency of these potential seizure events were shown to have a moderate to strong correlation with the PIP measures of stress. Correlation coefficients were 0.59 and 0.54 for the PIP-f and PIP-d scales respectively. This is concordant with previous research suggesting that seizure frequency is an influencing factor for caregiver stress (Austin and Caplan, 2007).

5.7.17 Statistics

Justifications for the statistical methods used throughout the results section of this thesis have been made in the discussions of the relevant sections. However, broadly speaking, the majority of the analyses involved non-parametric methods. This was done as we could not assume that the data was normally distributed. Evidenced by the histograms for the total sample conducted for the different questionnaires it was apparent that there was an element of skew to the overall data. This may be due partly to the recruitment approach, the small sample size and attritional bias (*i.e.* skew introduced based upon the nature of the people who remain in the study and those who do not). As a result of this calculations used for normally distributed data sets, such as means, standard deviations *etc.* were not appropriate for the majority of the results. This is not necessarily a limitation and indeed based upon the population targeted, carers of children with a suspected seizure disorder; it would not be possible to gain a normally distributed population.

5.7.18 Study Design Limitations

Sample Size

Overall, one of the most important limitations of this study was the small sample size. Despite best efforts, at the time of writing we obtained n=39 complete matched datasets. This has had several key effects. Primarily it affected the statistical methods available for the data analysis; this is discussed in greater detail later in this section, thereby limiting the amount of information obtainable from the data. It also may have the results in that a much greater effect size was necessary in order to have a statistically significant result. The small sample size may have additionally enhanced the effect of attritional bias. Attrition is expected for any longitudinal study, however having a smaller number at T0 means that any attrition will have a greater effect on the total population.

Time requirements and Number of Questionnaires

A major weakness of the study design and results is the small sample size and associated attrition regarding follow-ups. It is apparent that there were a number of potential barriers to recruitment which should be highlighted in order for alterations and/or improvements to be made in future work. In addition to recruitment barriers, the time requirements and volume of questionnaires also had some practical implications on the organisation of the study and the availability of a single researcher at any given time.

One of these perceived barriers is the length of time required to complete the questionnaires. The thirteen questionnaires used in this study each take up several pages worth of questions. As a result, the recruitment process was reasonably lengthy, taking on average between 30-45 minutes. This, in addition to a clinic appointment and a (probable) lengthy wait in the outpatient waiting rooms may have had a negative effect on recruitment.

In order to improve on this limitation and thus the strength of the findings, there are several potential adjustments that should be considered for future work in this area.

An alternative format for the study may be an option. Conducting the surveys using an online/ email form could aid recruitment numbers by having the option to complete the questionnaires from the comfort of their own home instead of a potentially busy and stressful hospital setting. Several secure sites exist and many medically oriented internet based audits have already been carried out, such as 'surveymonkey'. This approach may also have practical benefits for the researchers, namely data would already be computerised, thereby eliminating the need (and associated time involved) to manually transcribe participant responses onto the relevant databases and/or spreadsheets and limiting any potential human errors caused while entering

the data. Additionally, it would also dramatically reduce the printing and storage costs for this research. This design would however raise some issues of its own, such as: parents adequately understanding the questionnaires without a study researcher present, the home environment in which they complete the questionnaire influencing their scores *etc.*

Alteration of the questionnaire battery could also be considered. It should be evaluated whether 2 different measures both measuring stress are necessary for the questionnaire battery, particularly when the SSSQ from personal experience was the most time consuming of the questionnaires. Data collected from Rotter's LoC questionnaire for example, provided limited useful data in term of stress or significance. Exchange of this tool for a simpler and more health related LoC measure may be a potential adjustment. However, as previously emphasised, a variety of different questionnaires are necessary for the study. As previously detailed, in section 1.1.1, stress is well known to be a highly multi-factorial concept. The different dimension within a family amplifies this attribute. Therefore when seeking to gain an improved understanding of family stress associated with new paediatric epilepsy it is of definite importance to have wide number of measured covariates thus resulting in a wide number of questionnaires.

A flaw in the study which may also have affected the validity of the results lies in the statistics conducted in the study design phase of this research. No power calculation was carried out in preparation of the study going ahead. This means that there was no pre-determined number of participants that the study was aiming to recruit in order for the results to have sufficient statistical power. On discussing this with lead investigator and previous researchers it was clear that there was no similar study on which to base power calculations on and this was recognised as a potential weakness.

In practical terms, this will have had little effect on recruitment itself, obviously the aim still being to recruit the maximum number of participants possible. However without having a power calculation to refer back to it was more difficult to show true statistical significance in the results provided.

A further error is lack of clear hypotheses made prior to recruitment. The use of 11 different questionnaires means that an abundance of information was collected on the SPSS databases. The sheer volume of the data means that analysing it on its own without a specific question to be answered leaves it open to 'data dredging' or multiple testing bias (Kent, 2001). It is hoped that by early recognition of this potential source of error and by producing a set of null hypotheses and questions of interest prior to carrying out any data analysis that the results of the study are minimally affected by this form of bias.

A further potential limitation of this piece of research lies in the study design. Specifically the fact that although the purpose of the study, as stated in the original protocol, was to investigate variables affecting family stress in a paediatric epilepsy setting, the study itself was not designed around any particular models of family stress as with the work of Buelow and Austin among others (Pei-Fan, 2005, Buelow et al., 2006). This makes interpreting the results in relation to theory more complex. However, this aspect may be considered as both a strength as well as a limitation because it has been demonstrated that many different theories exist for the development of family stress and a plethora of factors have been implicated in its evolution. There is, as yet, no consensus on which, if any, of the theories is most plausible and therefore justified in the study design.

Questionnaires Selected and Statistics

The individual questionnaires themselves may also bring their own limitations which could, in turn, affect the study. A summary of each of these tools is presented in Section 5.5.5 Table 12.

Unfortunately, due to the small sample size of this study the original statistical method, using SEM, was not appropriate for analysing the results. Additionally, because there were less than 50 matched pairs for data, based on detailed discussions with statistical advisor Dr Lane, it was not possible to conduct valid change over time analyses. This would have been interesting in order to track how carers stress and other covariate measures changed throughout the study period from initial presentation to receiving a diagnosis, this could provide a greater level of information particularly regarding the epilepsy cohort.

This has been recognised as a weakness of the study as it limits the amount of results obtained from the collected data. Although it was not possible to conduct these analyses at the present time, it is hoped that with the ongoing T6 data collections a sample size >50 will be reached to allow for further valid statistical testing to enhance the information gained from this work.

Consistency of reporting in at 1st and 2nd meetings

An additional potential for bias is in the reporter consistency between the first meeting and the follow up meeting. The protocol stated the need of the questionnaires to be completed by the child's primary caregiver. In the event, the person accompanying the child to the outpatient appointment was the one who was approached for consent, which if given, received the first batch of questionnaires to be completed. At the follow up meeting the same system was used. However error may have been introduced as no record was kept of which parent/guardian completed the questionnaires. Individuals experience stress in different ways

and therefore if there was an inconsistency between which family members completed the questionnaires then this could potentially affect the reliability of the data.

After recognising this possible source of bias, efforts were made to limit this as much as possible. The first page of the SSSQ (See appendices) asks the responder to fill in their relation to the child in question. Paper copies for every participants' T0 and T1 SSSQ were searched in order to record onto the database the relation of the individual completing each batch of questionnaires. After noting this potential source of error, I attempted to limit it as much as possible. This was done when 2 carers arrived to clinic with their child I clarified which carer had completed the questionnaires the previous occasion and asked that carer to do so again. On at least 2 occasions a carer who had not completed the original set of questionnaires accompanied the child to clinic. In this situation to limit any source of error a set of questionnaires, instructions for completing questionnaires and a pre-addressed envelope were passed over to be given to the primary carer to send back.

Inter- operator variability

A potential limitation of this study is inter-operator variability. In the three years of its duration, this work has been carried out by three different M.Phil students. Although the questionnaires and study documentation remained the same throughout several aspects of the protocol were not necessarily explicit enough. An example of this appears in the selection criteria: it was specified that those children with other serious concomitant medical conditions be excluded from participation. However with no pre-determined definition of what would be classed as 'serious' there may be some variability regarding those recruited. Attempts were made to control this as much as possible by frequent interaction with the previous students to try and ensure continuity.

A further variability between operators may also have arisen during the selection process. When searching through the paediatric neurological referral letters there was no agreed upon terminology/phrases to look for in the letters that would point towards an epilepsy diagnosis. Therefore the selection of those patients to whom an information pack was sent to, was decided only based on the individual researchers opinion. While it is relevant to note that each investigator was an intercalating medical student (each of whom had already passed MBChB final examinations) and therefore could potentially be considered proficient in recognising a queried seizure disorder this may have caused a slight bias.

This potential source of error may be limited in the future by recruiting either a PhD student or other longer standing staff to complete such work to ensure continuity and minimise observer bias.

Study Location

Another potential, although inevitable, limitation of this study is its location: Alder Hey Children's Hospital. It is one of few paediatric tertiary care centres in the North West and the U.K as a whole. This setting may affect the ability to apply these findings to a general population. It is conceivable that coming to an appointment at a large and famous tertiary care hospital may be more stressful than attending either a general practice appointment or a clinic within a smaller local district general hospital.

Types of Epilepsy

Broadly speaking, the division of cohorts into those participants with epilepsy and those without serves as an adequate distinction for the study question, there are a few points related to this categorisation which could be improved. Information was only collected upon the broad category of seizure type i.e. focal or generalised morphology. Different seizures may potentially cause a greater degree of stress, for example comparing a generalised tonic-clonic seizure to an absence seizure. Different seizures are attributable to different causes of epilepsy, see classification section in Chapter 2. Therefore further information collected on the nature of the seizures/events and the syndromic epilepsy classification might provide more detailed information regarding stress in epilepsy in future research. Unfortunately even if collected it would be of limited import to this study due to the small sample size, meaning that subdivisions within the epilepsy cohort would likely be too small to carry out adequate statistical analysis. However this idea could be carried forward in any future larger scale work.

5.7.18 Additional Ideas for Further Work

A potential direction for further work following on from this study would be the development of an interventional based study regarding carer stress and paediatric epilepsy. Before deciding what form of intervention would be most appropriate- by taking into consideration parental needs but also practical issues such as a cost-benefit analysis, it would be important to audit the existing interventions available to these families. Examples of these include the epilepsy nurses, the 'first fit' pamphlets, carer psychology referrals. Evaluation of these services may bring to attention any adaptations or adjustments which may improve their utility and efficacy.

Associated with future interventional work, development of a screening tool such as an abbreviation of this study's questionnaire battery should be considered. In the current economic climate interventional services may not be available for every single family therefore identifying those carer's at greater risk of reaching a crisis is of value. In order to adequately assess this a pilot of greater numbers, for example n=50 matched, would be required at both time points to allow for statistically valid regression analyses or SEM

techniques to determine which would be the most useful factors (and therefore, questionnaires) to focus on for such a screening tool.

The impact of fatigue and its affect on carer stress could be a factor of interest within the context of paediatric epilepsy. Existing research has shown positive associations between carer fatigue and stress (Clark, 2002). This work has mainly been done on carers of adults with dementia, but it would be interesting to understand if this finding is also replicated in carers of children with chronic problems and epilepsy in particular. However this variable does have limitations, primarily with regard to potential of planning effective interventions should such a correlation be found.

A further direction for future research in this field could involve surveying the siblings of children with epilepsy to also complete appropriate questionnaires- not only about their affected brother or sister but also fill in their perceptions about their own stress levels. This could provide interesting information, as it could potentially be hypothesised that while the parents/carers focus more upon the affected child the 'normal' sibling may feel resentful and stressed. This could lead to a negative impact on the unaffected child also. This is an idea that has previously been identified in other fields, primarily family functioning in paediatric psychology (Sharpe and Rossiter, 2002). As a result specific questionnaires have already been developed for siblings. One example of which is the Family Life Questionnaire (FaLQ), which is a brief 11 item tool, so in practical terms of additional workload and time constraints it has potential for use in a similarly designed study (Sharpe and Rossiter, 2002).

Another interesting concept which has risen to prominence is that of emotional intelligence. It can be simply defined as an individuals' ability to identify, evaluate and control the emotions of both themselves and those of others (Houghton et al., 2012). Prior research has already shown an association between stress and emotional intelligence in samples of students (Houghton et al., 2012). Therefore it may prove interesting to determine whether a parent or carer's emotional intelligence has an impact on their perceived stress levels amongst other covariates.

5.8 Overall Discussion and Conclusions

This study adds to the existing knowledge base surrounding carer stress when a child is newly diagnosed with epilepsy. This was done by building upon the understanding of how carer stress levels change within the first 6 months from first presentation and diagnosis. At the time of writing, this author is aware of only 5 other studies investigating stress at the time of new diagnosis of paediatric epilepsy.

Following on from a pilot study carried in August 2010, this study measured 7 other covariates in addition to stress itself, in order to reflect the multi-factorial nature of the concept and assess these variables within a paediatric epilepsy context. These covariates included: carer health, coping methods, support, needs, locus of control, family functioning, family communication, child quality of life, child self-image and child behaviour. The novel aspect of using a control group of children (and their parents/carers) who have also experienced the same pathway and uncertainty regarding a potential diagnosis of epilepsy seeks to clarify our current knowledge of the development of stress in this group of carers.

The overall hypotheses of this study were:

1. There is no difference in carer stress scores between cohorts at T0

Following analysis of the data collected by this study and the resulting statistical analyses it is clear that we should continue to accept the null hypothesis. None of the questionnaires found any statistically significant differences in stress or any of its associated covariates between the 2 cohorts at T0. This indicates that at time of presentation and prior to receiving any diagnosis there are no differences between the stress experienced by carers regardless of whether their child is later confirmed to have epilepsy.

In some ways this was the expected finding. Both carers have gone through similar experiences up until this point, namely their child having had suspected seizure events, the referral process and the wait for a scheduled clinic appointment at AHCH. However most interestingly, at their first presentation although there were no significant differences between the 2 groups 59% of all carers regardless of their cohort reported greater than normal stress scores. In contrast with some of the previous work by Modi found no difference between the parental stress of those whose children had new onset epilepsy and the healthy controls, where only 7% of carers were experiencing higher levels of stress (Modi, 2009). Such differences may partly be down to the different outcome measures used having different levels of specificity and due to cultural/geographical differences between the study populations. However, it still emphasises that evaluation of existing supports in places at AHCH may be of use and provides a suitable starting point for any future interventional work.

2. There is no difference in carer stress scores between cohorts at T6

As described in the analysis, the results of this study found statistically significant differences between the stress scores of the epilepsy and non epilepsy cohorts at T6. This was observed in the 2 different measures of stress utilised by this research, the PIP and the SSSQ.

These findings suggest that we may reject the null hypothesis and that a clear difference in carer stress scores at the second data collection point was demonstrated in this work. This agrees with previous research findings such as that by Chiou describing that carers whose children received a diagnosis of epilepsy exhibited high stress levels and those levels exceeded the stress of carers whose children were diagnosed with asthma (Chiou and Hsieh, 2008). However, a limitation common with some of this previous work that it is impossible to directly compare one disease with another. To date this author knows of no other study using a control group in which the carers have also experienced the stress of a potential paediatric epilepsy disorder. By determining a significant difference between who had received an epilepsy diagnosis compared to those who had not, when there had been no previous difference between the cohorts at initial presentation suggests that it may not necessarily be the disease itself causing stress but rather the parent's reaction to the diagnosis itself and their preconceived ideas about its consequences.

It may also imply that the diagnosis itself, not just the condition is also a variable influencing a child's view of their quality of life. Previous work that reported decreased quality of life in children with epilepsy has often used a sample where the diagnosis was established for a greater period of time. In our study these children reported no significantly different quality of life scores compared to the control, however, having only just received this information, it possible that they have not yet experienced how this diagnosis may alter their lives in a practical setting. It would be naive to assume that this would be the only factor- medication, social life restriction and school among others would also logically be implicated in the child's quality of life. It would be interesting to follow up any changes in the children's perceived quality of life after they have had a chance to introduce their epilepsy and its implications into context with their daily lives.

3. There is no association between carer stress and the other variables

All of the covariates investigated in this study were chosen due to their demonstrated association with stress either within family stress theory or in the existing literature. Therefore, when correlations were conducted between the covariate total scores and stress total scores collected from this sample it was fascinating to observe the different levels of association between these.

The strongest correlations with stress were found with child quality of life, child behaviour and carer health. It is noted that two of these three covariates are child oriented. This may be intuitive given the ideal that parents and carers put their child's needs before their own, and family systems theories' indicate the strong interconnecting links with different family members. However, considering the basic principles it is interesting to note that external

variables i.e. those affecting another person were more highly correlated to stress than personal or internal variables such as the carer's perceived needs.

This also leads on to one of the more unexpected findings- the absence of strong correlation between carer stress and total carer support. This was especially surprising given the existing knowledge base of support, be it familial or social, being implicated as a strong mediating factor regarding stress. It is unclear how those families' experiencing stress despite having high levels of support would be managing without support and whether this support prevented them from descending into crisis. This may link with family resilience and the individuality of each family as described in Chapter 1.

Although no strong correlations were found between the demographic variables, those most implicated were carer age, level of carer education and number of children in the family home. Also, unsurprising, the frequency of the potentially epileptic events were found to have a moderate to strong correlation with carer stress. This is logical and in keeping with existing knowledge. Despite this, it was noted that the strongest correlations with stress were found by this demographic variable rather than the associated covariates. This suggests for future screening of carer's who may be in need of assistance or interventions regarding dealing with stress the inclusion of carer demographics and seizure characteristics should not be overlooked.

Overall, the existing knowledge base consistently recognises the association of paediatric epilepsy with increased carer stress. However, a paucity of literature and need of greater investigation into the carer stress levels at the onset and diagnosis of new paediatric epilepsy has been acknowledged. This study has aimed to help improve the understanding within this area by researching not only stress but some of its covariates over time. The results suggest that at first presentation to secondary care, regardless of their child's diagnosis the majority of carers (59%) are experiencing increased levels of stress. It is hard to differentiate how much of this is accounted for by just having to attend a hospital, potentially for the first time, or the suspected epilepsy. However, approximately 6 months later, after diagnosis the stress levels of carers of children with epilepsy remain high and are significantly greater than those carers in the non-epilepsy cohort. Not only do the carers of the epilepsy group exhibit greater stress but also greater mental health symptoms, increased levels of dysfunctional coping strategies and increased difficult child behaviour concerns.

Based upon the nature of the cohorts, in that the non-epilepsy group have been through the same healthcare pathway, it may be suggested that the uncertainty and the care pathway itself may be a stressor for carers coming to Alder Hey Children's Hospital for a new diagnosis. This may be important information in evaluating the hospital procedure and lead to an audit of methods by which carer stress levels may be minimised. Additionally, it indicates

that receiving the diagnosis of epilepsy may cause similar degrees of stress (if not more) than the seizures themselves based upon the significant increase in stress between cohorts following diagnosis, rather than throughout the process. This raises an interesting prospect in need of greater elucidation. The higher correlations between stress and the variables of child quality of life, child behaviour and carer health provide a broader understanding and may also identify specific areas in which an intervention may be targeted to help reduce the stress and improve the wellbeing of these families.

References

- APPLETON, R. G., J. 2004. Epilepsy in Childhood and Adolescence. *Epilepsy in Childhood and Adolescence*. 3rd Edition ed. Informa UK Limited: Taylor and Francis.
- APPLETON, R. M., ANTONY 2009a. Living with Epilepsy. *Epilepsy: the facts*. 3rd Edition ed.: Oxford University Press.
- APPLETON, R. M., ANTONY 2009b. The treatment of epilepsy. *Epilepsy: the facts*. 3rd Edition ed.: Oxford University Press.
- AUER, R. N. & SIESJO, B. K. 1988. Biological differences between ischemia, hypoglycemia, and epilepsy. *Annals of Neurology*, 24, 699-707.
- AUSTIN, J. K. & CAPLAN, R. 2007. Behavioral and Psychiatric Comorbidities in Pediatric Epilepsy: Toward an Integrative Model. *Epilepsia*, 48, 1639-1651.
- AUSTIN, J. K., HAREZLAK, J., DUNN, D. W., HUSTER, G. A., ROSE, D. F. & AMBROSIUS, W. T. 2001. Behavior Problems in Children Before First Recognized Seizures. *Pediatrics*, 107, 115-122.
- BACA, C. B., VICKREY, B. G., CAPLAN, R., VASSAR, S. D. & BERG, A. T. 2011. Psychiatric and Medical Comorbidity and Quality of Life Outcomes in Childhood-Onset Epilepsy. *Pediatrics*, 128, e1532-e1543.
- BACA, C. B., VICKREY, B. G., HAYS, R. D., VASSAR, S. D. & BERG, A. T. 2010. Differences in Child versus Parent Reports of the Child's Health-Related Quality of Life in Children with Epilepsy and Healthy Siblings. *Value in Health*, 13, 778-786.
- BAILEY, D. B. & SIMEONSSON, R. J. 1988. Assessing Needs of Families with Handicapped Infants. *The Journal of Special Education*, 22, 117-127.
- BAKER, G. 2011. Epilepsy in Childhood: A Companions guide to Epilepsy. In: ROGAN, P. (ed.). Sanofi-Synthelabo.
- BANERJEE, P. N., FILIPPI, D. & ALLEN HAUSER, W. 2009. The descriptive epidemiology of epilepsy—A review. *Epilepsy Research*, 85, 31-45.
- BARAKAT, L., LUTZ, M., NICOLAOU, D. & LASH, L. 2005. Parental Locus of Control and Family Functioning in the Quality of Life of Children with Sickle Cell Disease. *Journal of Clinical Psychology in Medical Settings*, 12, 323-331.
- BAUMANN, A. T., F. 2010. Definition of Stress, stressor, eustress [Online]. Available: <http://www.stress.org/topic-definition-stress.htm> [Accessed 14/07/2011 2011].
- BAUMANN, N. & TURPIN, J. C. 2010. Neurochemistry of stress. An overview. *Neurochem Res*, 35, 1875-9.
- BENGTON, V. L. 2001. Beyond the Nuclear Family: The Increasing Importance of Multigenerational Bonds. *Journal of Marriage and Family*, 63, 1-16.
- BERETVAS, S. N., SUIZZO, M.-A., DURHAM, J. A. & YARNELL, L. M. 2008. A Reliability Generalization Study of Scores on Rotter's and Nowicki-Strickland's Locus of Control Scales. *Educational and Psychological Measurement*, 68, 97-119.
- BERG, A. 2001. Mortality in Epilepsy. *Epilepsy Currents*, 1, 28-28.
- BERG, A. T., BERKOVIC, S. F., BRODIE, M. J., BUCHHALTER, J., CROSS, J. H., VAN EMDE BOAS, W., ENGEL, J., FRENCH, J., GLAUSER, T. A., MATHERN, G. W., MOSHÉ, S. L., NORDLI, D., PLOUIN, P. & SCHEFFER, I. E. 2010. Revised terminology and concepts for organization of seizures and epilepsies: Report of the ILAE Commission on Classification and Terminology, 2005-2009. *Epilepsia*, 51, 676-685.
- BERG, A. T. & SCHEFFER, I. E. 2011. New concepts in classification of the epilepsies: Entering the 21st century. *Epilepsia*, 52, 1058-1062.
- BILLINGS, A. G. & MOOS, R. H. 1981. The role of coping responses and social resources in attenuating the stress of life events. *Journal of Behavioral Medicine*, 4, 139-157.

- BOSS, P. 2002. *Family Stress Management: a contextual approach*, London, EC2A 4PU, U.k., SAGE Publications Inc.
- BOSS, P. & GREENBERG, J. 1984. Family boundary ambiguity: a new variable in family stress theory. *Fam Process*, 23, 535-46.
- BOUMA, R. & SCHWEITZER, R. 1990. The impact of chronic childhood illness on family stress: A comparison between autism and cystic fibrosis. *Journal of Clinical Psychology*, 46, 722-730.
- BOWLING, A. 2009. *Research Methods in Health*, Open University Press.
- BUELOW, J. M., MCNELIS, A., SHORE, C. P. & AUSTIN, J. K. 2006. Stressors of parents of children with epilepsy and intellectual disability. *Journal of Neuroscience Nursing*, 38, 147.
- BURGESS, E. D., D; TAYLOR, GERRY; WADE, SHARI; STANCIN, TERRY. 2008. The Family Burden of Injury Interview: Reliability and Validity Scales. *Journal of Head Trauma Rehabilitation*, 14, 394-405.
- BUTLER, R. 2001. The Self Image Profiles For Children (SIP-C) and Adolescents (SIP-A) Manual. Pearson Ltd.
- BUTLER, R. J. & GASSON, S. L. 2005. Self Esteem/Self Concept Scales for Children and Adolescents: A Review. *Child and Adolescent Mental Health*, 10, 190-201.
- CAMFIELD, P., CAMFIELD, C. & NOLAN, K. Helping families cope with the devastation of Dravet syndrome. *European Journal of Paediatric Neurology*.
- CARVER, C. S. 1997. You want to Measure Coping But Your Protocol's Too Long: Consider the Brief COPE. *International Journal of Behavioral Medicine*, 4, 92.
- CARVER, C. S. 2007. *Brief COPE* [Online]. University of Miami. Available: <http://www.psy.miami.edu/faculty/ccarver/sclBrCOPE.html> [Accessed 3/2/12.
- CHIOU, H.-H. & HSIEH, L.-P. 2008. Parenting Stress in Parents of Children With Epilepsy and Asthma. *Journal of Child Neurology*, 23, 301-306.
- CLARK, P. C. 2002. Effects of individual and family hardiness on caregiver depression and fatigue†. *Research in Nursing & Health*, 25, 37-48.
- COOPER, C., KATONA, C. & LIVINGSTON, G. 2008. Validity and Reliability of the Brief COPE in Carers of People With Dementia: The LASER-AD Study. *The Journal of Nervous and Mental Disease*, 196, 838-843
10.1097/NMD.0b013e31818b504c.
- COUNCIL, G. M. 2010. Good practice in research and consent to research. 3 ed.: GMC.
- COX, M. J. & PALEY, B. 1997. Families as system. *Annual Review of Psychology*, 48, 243.
- CRAWFORD, P. 2005. Best Practice Guidelines for the Management of Women with Epilepsy. *Epilepsia*, 46, 117-124.
- DAVIS, S. E., HYNAN, L. S., LIMBERS, C. A., ANDERSEN, C. M., GREENE, M. C., VARNI, J. W. & IANNACCONE, S. T. 2010. The PedsQL™ in pediatric patients with duchenne muscular dystrophy: Feasibility, reliability, and validity of the pediatric quality of life inventory neuromuscular module and generic core scales. *Journal of Clinical Neuromuscular Disease*, 11, 97-109.
- DE BOER, H. M. 2010. Epilepsy stigma: Moving from a global problem to global solutions. *Seizure*, 19, 630-636.
- DEVINSKY, O. 2011. Sudden, Unexpected Death in Epilepsy. *New England Journal of Medicine*, 365, 1801-1811.
- DEVINSKY, O., WESTBROOK, L., CRAMER, J., GLASSMAN, M., PERRINE, K. & CAMFIELD, C. 1999. Risk factors for poor health-related quality of life in adolescents with epilepsy. *Epilepsia*, 40, 1715-1720.
- DIAMANTIS, A., SIDIROPOULOU, K. & MAGIORKINIS, E. 2010. Epilepsy during the Middle Ages, the Renaissance and the Enlightenment. *J Neurol.*, 257, 691-8.

- DONG, B. E. A. 2011. *The Theory of Family Stress and Adaptation* [Online]. Available: <http://www.plu.edu/~hopeweeh/doc/family-stress.ppt> [Accessed 23/07/2011 2011].
- DONNELLAN, C., HEVEY, D., HICKEY, A. & O'NEILL, D. 2006. Defining and quantifying coping strategies after stroke: a review. *Journal of Neurology, Neurosurgery & Psychiatry*, 77, 1208-1218.
- DUFFY, L. V. 2011. Parental coping and childhood epilepsy: the need for future research. *J Neurosci Nurs*, 43, 29-35.
- DUNST, C. J. & TRIVETTE, C. M. 1986. Mediating influences of social support: Personal, family, and child outcomes. *American Journal of Mental Deficiency*, 90, 403-417.
- EKINCI, O., TITUS, J. B., RODOPMAN, A. A., BERKEM, M. & TREVATHAN, E. 2009. Depression and anxiety in children and adolescents with epilepsy: Prevalence, risk factors, and treatment. *Epilepsy & Behavior*, 14, 8-18.
- EPILEPSYACTION. 2011a. *Driving Agencies and Licence Categories* [Online]. Epilepsy Action. Available: <http://www.epilepsy.org.uk/info/driving/agencies-licence-categories#standards> [2012].
- EPILEPSYACTION. 2011b. *Supporting your Child with Epilepsy* [Online]. Available: <http://www.epilepsy.org.uk/info/children/children-with-epilepsy/supporting> [Accessed 17/6/12].
- F, B. & S, N. 2010. PARENTAL KNOWLEDGE AND MENTAL HEALTH IN PARENTS OF CHILDREN WITH EPILEPSY. *Pakistan Journal of Medical Sciences*, 26, 191.
- FAILDE, I., RAMOS, I. & FERNANDEZ-PALACÍN, F. 2000. Comparison between the GHQ-28 and SF-36 (MH 1-5) for the assessment of the mental health in patients with ischaemic heart disease. *European Journal of Epidemiology*, 16, 311-316.
- FARMER, J. E., MARIEN, W. E., CLARK, M. J., SHERMAN, A. & SELVA, T. J. 2004. Primary Care Supports for Children with Chronic Health Conditions: Identifying and Predicting Unmet Family Needs. *Journal of Pediatric Psychology*, 29, 355-367.
- FERRO, M., AVISON, W., CAMPBELL, M. & SPEECHLEY, K. 2010. Do depressive symptoms affect mothers' reports of child outcomes in children with new-onset epilepsy? *Quality of Life Research*, 19, 955-964.
- FRIEDRICH, F., ALEXANDROWICZ, R., BENDA, N., CERNY, G. & WANCATA, J. 2011. The criterion validity of different versions of the General Health Questionnaire among non-psychiatric inpatients. *Social Psychiatry And Psychiatric Epidemiology*, 46, 635-641.
- GAUDET, L. P. G. 1989. Systems treatment in pediatric chronic illness: a parent group program. *Family Systems Medicine*, 7, 90-99.
- GAUTHIER-WEBER, J. 2011. *Individual and Family Stress and Crises*, Sage.
- GOODMAN, R. 1997. The Strengths and Difficulties Questionnaire: A Research Note. *Journal of Child Psychology and Psychiatry*, 38, 581-586.
- GOODMAN, R. 2001. Psychometric properties of the strengths and difficulties questionnaire. *Journal of the American Academy of Child and Adolescent Psychiatry*, 40, 1337-1345.
- GOODMAN, R. & SCOTT, S. 1999. Comparing the Strengths and Difficulties Questionnaire and the Child Behavior Checklist: is small beautiful? *J Abnorm Child Psychol*, 27, 17-24.
- GREEN, J. T., N. 2011. What is Qualitative Research? *Qualitative Methods for Health Research*. Second Edition ed.: SAGE Publications Ltd.
- HALL, H. R., NEELY-BARNES, S. L., GRAFF, J. C., KRCEK, T. E., ROBERTS, R. J. & HANKINS, J. S. 2012. Parental Stress in Families of Children with a Genetic Disorder/Disability and the Resiliency Model of Family Stress,

- Adjustment, and Adaptation. *Issues in Comprehensive Pediatric Nursing*, 35, 24-44.
- HANLEY, B., TASSÉ, M. J., AMAN, M. G. & PACE, P. 1998. Psychometric Properties of the Family Support Scale with Head Start Families. *Journal of Child and Family Studies*, 7, 69-77.
- HASKETT, M. E., AHERN, L. S., WARD, C. S. & ALLAIRE, J. C. 2006. Factor Structure and Validity of the Parenting Stress Index-Short Form. *Journal of Clinical Child and Adolescent Psychology*, 35, 302-312.
- HAWLEY, D. R. & DEHAAN, L. 1996. Toward a Definition of Family Resilience: Integrating Life-Span and Family Perspectives. *Family Process*, 35, 283-298.
- HEALTH), S. S. F. P. 2012. *Critical Appraisal Skills Programme* [Online]. NHS. Available: <http://www.sph.nhs.uk/what-we-do/public-health-workforce/resources/critical-appraisals-skills-programme> 2012].
- HOCH, D. B. 2010. Cerebral Hypoxia. In: HEALTH, N. I. O. (ed.) *Medline Plus*. U.S. National Library of Medicine.
- HOLMES, G. L. & BEN-ARI, Y. 2001. The Neurobiology and Consequences of Epilepsy in the Developing Brain. *Pediatr Res*, 49, 320-325.
- HOLROYD, J. & GUTHRIE, D. 1986. Family stress with chronic childhood illness: Cystic fibrosis, neuromuscular disease, and renal disease. *Journal of Clinical Psychology*, 42, 552-561.
- HOME, P. & KERIREY, S. 1991. PSYCHOSOCIAL ADJUSTMENT OF CHILDREN WITH CHRONIC EPILEPSY AND THEIR FAMILIES. *Developmental Medicine & Child Neurology*, 33, 201-215.
- HOUGHTON, J. D., WU, J., GODWIN, J. L., NECK, C. P. & MANZ, C. C. 2012. Effective Stress Management. *Journal of Management Education*, 36, 220-238.
- HUA-HUEI, C. & LIANG-PO, H. 2008. Comparative Study of Children's Self-Concepts and Parenting Stress Between Families of Children With Epilepsy and Asthma. *Journal of Nursing Research (Taiwan Nurses Association)*, 16, 65-73.
- KENT, R. 2001. Analysing survey data: knowing how to handle your data. *Data Construction and Data analysis For Survey Research*. 1 ed.: Palgrave Publishing Inc.
- KLIMACH, V. J. 2009. The community use of rescue medication for prolonged epileptic seizures in children. *Seizure : the journal of the British Epilepsy Association*, 18, 343-346.
- KORMANIK, M. B. & ROCCO, T. S. 2009. Internal versus external control of reinforcement: A review of the locus of control construct. *Human Resource Development Review*, 8, 463-483.
- KRATZ, L., UDING, N., TRAHMS, C. M., VILLAREALE, N. & KIECKHEFER, G. M. 2009. Managing childhood chronic illness: Parent perspectives and implications for parent-provider relationships. *Families, Systems, & Health*, 27, 303-313.
- LAVEE, Y. M., H. PATTERSON, J 1985. The Double ABCX Model of Family Stress and Adaptation: An Empirical Test by Analysis of Structural Equations with Latent Variables. *Journal of Marriage and Family*, 47, 811-825.
- LEE, P. 2010. Has disability discrimination legislation changed the legal framework for epilepsy in the United Kingdom? *Seizure*, 19, 619-622.
- LEVY, J. R., A. BOTUCK, S. ARDITO, M 1996. The support network of mothers of younger and adult children with mental retardation and developmental disabilities receiving care. *British Journal of Developmental Disabilities*, XL11, 82.
- LOYD, B. H. & ABIDIN, R. R. 1985. Revision of the Parenting Stress Index. *Journal of Pediatric Psychology*, 10, 169-177.

- LUSZCZYNSKA, A. & SCHWARZER, R. 2005. Multidimensional Health Locus of Control: Comments on the construct and its measurement. *Journal of Health Psychology*, 10, 633-642.
- MAGIORKINIS, E., SIDIROPOULOU, K. & DIAMANTIS, A. 2010. Hallmarks in the history of epilepsy: Epilepsy in antiquity. *Epilepsy & Behavior*, 17, 103-108.
- MARSAC, M. L. & ALDERFER, M. A. 2011. Psychometric Properties of the FACES-IV in a Pediatric Oncology Population. *Journal of Pediatric Psychology*, 36, 528-538.
- MARSON, A. G., AL-KHARUSI, A. M., ALWAIDH, M., APPLETON, R., BAKER, G. A., CHADWICK, D. W., CRAMP, C., COCKERELL, O. C., COOPER, P. N., DOUGHTY, J., EATON, B., GAMBLE, C., GOULDING, P. J., HOWELL, S. J. L., HUGHES, A., JACKSON, M., JACOBY, A., KELLETT, M., LAWSON, G. R., LEACH, J. P., NICOLAIDES, P., ROBERTS, R., SHACKLEY, P., SHEN, J., SMITH, D. F., SMITH, P. E. M., SMITH, C. T., VANOLI, A. & WILLIAMSON, P. R. 2007. The SANAD study of effectiveness of carbamazepine, gabapentin, lamotrigine, oxcarbazepine, or topiramate for treatment of partial epilepsy: an unblinded randomised controlled trial. *The Lancet*, 369, 1000-1015.
- MAYS, R. M. 1988. Family stress and adaptation. *Nurse Pract*, 13, 52, 54, 56.
- MCCUBBIN, H. I. 1979. Integrating Coping Behavior in Family Stress Theory. *Journal of Marriage and the Family*, 41, 237-244.
- MCCUBBIN, H. I., JOY, C. B., CAUBLE, A. E., COMEAU, J. K., PATTERSON, J. M. & NEEDLE, R. H. 1980. Family Stress and Coping - a Decade Review. *Journal of Marriage and the Family*, 42, 855-871.
- MCTAGUE, A. & APPLETON, R. 2011. Treatment of difficult epilepsy. *Archives of Disease in Childhood*, 96, 200-204.
- MEDNICK, L., GARGOLLO, P., OLIVA, M., GRANT, R. & BORER, J. 2009. Stress and Coping of Parents of Young Children Diagnosed With Bladder Exstrophy. *The Journal of Urology*, 181, 1312-1317.
- MILTON, J. G. 2010. Epilepsy as a dynamic disease: A tutorial of the past with an eye to the future. *Epilepsy and Behavior*, 18, 33-44.
- MIND, Y. I. 2011. *SDQ Info: Youth In Mind* [Online]. Available: <http://www.sdqinfo.org/g0.html>.
- MINDRUTA, I., DIMITRIU, A. & CRACIUN, L. 2011. Insights in classification of seizures and epilepsies-review of key concepts throughout history. *Romanian Journal of Neurology/ Revista Romana de Neurologie*, 10, 161-171.
- MODI, A. C. 2009. The impact of a new pediatric epilepsy diagnosis on parents: Parenting stress and activity patterns. *Epilepsy & Behavior*, 14, 237-242.
- MOHER, D., COOK, D. J., EASTWOOD, S., OLKIN, I., RENNIE, D. & STROUP, D. F. 1999. Improving the quality of reports of meta-analyses of randomised controlled trials: the QUOROM statement. Quality of Reporting of Meta-analyses. *Lancet*, 354, 1896-1900.
- MORRELL, M. J. 2002. Stigma and epilepsy. *Epilepsy & Behavior*, 3, 21-25.
- MU, P.-F., KUO, H.-C. & CHANG, K.-P. 2005. Boundary ambiguity, coping patterns and depression in mothers caring for children with epilepsy in Taiwan. *International Journal of Nursing Studies*, 42, 273-282.
- MURRAY, J. 1993. Coping with the uncertainty of uncontrolled epilepsy. *Seizure*, 2, 167-178.
- NASHEF, L., SO, E. L., RYVLIN, P. & TOMSON, T. 2012. Unifying the definitions of sudden unexpected death in epilepsy. *Epilepsia*, 53, 227-233.
- NEGRAO, A. B., DEUSTER, P. A., GOLD, P. W., SINGH, A. & CHROUSOS, G. P. 2000. Individual reactivity and physiology of the stress response. *Biomed Pharmacother*, 54, 122-8.

- NICE 2012a. CG137 Appendix D: Differential Diagnosis of Epilepsy in Adults and Children. *The epilepsies: the diagnosis and management of epilepsies in adults and children*.
- NICE 2012b. The epilepsies: the diagnosis and management of the epilepsies in adults and children in primary and secondary care. *NICE Clinical Guidelines*, 137, 7-27.
- NICE 2012c. The epilepsies: the diagnosis and management of the epilepsies in adults and children in primary and secondary care. NICE.
- NICE 2012d. The epilepsies: the diagnosis and management of the epilepsies in adults and children in primary and secondary care. NICE.
- NIKOLAS, H., RAJIV, M., JOHN, N. & MARTIN, J. B. 2007. Review: Mortality in epilepsy. *Epilepsy and Behavior*, 10, 363-376.
- OGDEN, J. 2007. *Health Psychology: A textbook*, McGraw Hill Ltd.
- OHLEYER, V., FREDDO, M., BAGNER, D. M., SIMONS, L. E., GEFFKEN, G. R., SILVERSTEIN, J. H. & STORCH, E. A. 2007. Disease-related stress in parents of children who are overweight: relations with parental anxiety and childhood psychosocial functioning. *Journal of Child Health Care*, 11, 132-142.
- OLIVERI, D. R. M. 1980. Family Paradigm and Family Coping: A Proposal for Linking the Family's Intrinsic Adaptive Capacities To Its Responses To Stress. *Family Relations*, 29, 431-444.
- OLSON, D. 2010. *FACES IV: Data Analysis Using FACES IV Scores* [Online]. Life Innovations. Available: www.faceiv.com/pdf/data_analysis.pdf [Accessed 04/06/2012 2012].
- OLSON, D. 2011. FACES IV and the Circumplex Model: Validation Study. *Journal of Marital and Family Therapy*, 37, 64-80.
- OLSON, D. H. 1988. Circumplex model of family systems VIII: Family assessment and intervention. *Journal of Psychotherapy and The Family*, 4, 7-49.
- OLSON, D. H., SPRENKLE, D. H. & RUSSELL, C. S. 1979. Circumplex Model of Marital and Family Systems: I. Cohesion and Adaptability Dimensions, Family Types, and Clinical Applications. *Family Process*, 18, 3-28.
- PANAYIOTOPOULOS, C. P. 2012. The new ILAE report on terminology and concepts for the organization of epilepsies: Critical review and contribution. *Epilepsia*.
- PATTERSON, J. M. 1988. Families experiencing stress: I. The Family Adjustment and Adaptation Response Model: II. Applying the FAAR Model to health-related issues for intervention and research. *Family Systems Medicine*, 6, 202-237.
- PATTERSON, J. M. & GARWICK, A. W. 1994. Levels of meaning in family stress theory. *Fam Process*, 33, 287-304.
- PEI-FAN, M. 2005. Paternal reactions to a child with epilepsy: uncertainty, coping strategies, and depression. *Journal of Advanced Nursing*, 49, 367-376.
- PERRIN, E. C. & SHAPIRO, E. 1985. Health locus of control beliefs of healthy children, children with a chronic physical illness, and their mothers. *The Journal Of Pediatrics*, 107, 627-633.
- PIT-TEN CATE, I. M., KENNEDY, C. & STEVENSON, J. 2002. Disability and quality of life in spina bifida and hydrocephalus. *Developmental Medicine & Child Neurology*, 44, 317-322.
- PLANT, K. M. & SANDERS, M. R. 2007. Predictors of care-giver stress in families of preschool-aged children with developmental disabilities. *Journal of Intellectual Disability Research*, 51, 109-124.
- PRESS, O. U. 2007. The Oxford Concise Medical Dictionary. In: MARTIN, E. (ed.) *The Oxford Concise Medical Dictionary*. 7th Edition ed.: Oxford University Press.

- RAINA, P., O'DONNELL, M., SCHWELLNUS, H., ROSENBAUM, P., KING, G., BREHAUT, J., RUSSELL, D., SWINTON, M., KING, S., WONG, M., WALTER, S. D. & WOOD, E. 2004. Caregiving process and caregiver burden: conceptual models to guide research and practice. *BMC Pediatrics*, 4, 1-1.
- RANGE, L. M. & STRINGER, T. A. 1996. Reasons for Living and Coping Abilities among Older Adults. *International Journal of Aging & Human Development*, 43, 1-5.
- REITMAN, D., CURRIER, R. O. & STICKLE, T. R. 2002. A critical evaluation of the Parenting Stress Index-Short Form (PSI-SF) in a head start population. *Journal Of Clinical Child And Adolescent Psychology: The Official Journal For The Society Of Clinical Child And Adolescent Psychology, American Psychological Association, Division 53*, 31, 384-392.
- RODENBURG, R., MEIJER, A. M., DEKOVIĆ, M. & ALDENKAMP, A. P. 2005. Family factors and psychopathology in children with epilepsy: A literature review. *Epilepsy & Behavior*, 6, 488-503.
- RODENBURG, R., MEIJER, A. M., DEKOVIĆ, M. & ALDENKAMP, A. P. 2007. Parents of children with enduring epilepsy: Predictors of parenting stress and parenting. *Epilepsy & Behavior*, 11, 197-207.
- RODENBURG, R., WAGNER, J. L., AUSTIN, J. K., KERR, M. & DUNN, D. W. 2011. Psychosocial issues for children with epilepsy. *Epilepsy & Behavior*, 22, 47-54.
- ROSCH, P. 2011. *Hans Selye* [Online]. Available: <http://www.stress.org/hans.htm> [Accessed 10/07/2011 2011].
- ROTTER, J. B. 1966. Generalized expectancies for internal versus external control of reinforcement. *Psychological Monographs: General and Applied*, 80, 1-28.
- RUTTER, M., TIZARD, J., YULE, W., GRAHAM, P. & WHITMORE, K. 1976. Isle of Wight Studies, 1964–1974. *Psychological Medicine*, 6, 313-332.
- SAMUEL-HODGE, C. D., WATKINS, D. C., ROWELL, K. L. & HOOTEN, E. G. 2008. Coping styles, well-being, and self-care behaviors among African Americans with type 2 diabetes. *Diabetes Educ*, 34, 501-10.
- SANDER, J. W. 2003. The epidemiology of epilepsy revisited. *Current Opinion in Neurology*, 16, 165-170.
- SANDER, J. W. A. S., HART, Y. M., SHORVON, S. D. & JOHNSON, A. L. 1990. MEDICAL SCIENCE: National General Practice Study of Epilepsy: newly diagnosed epileptic seizures in a general population. *The Lancet*, 336, 1267-1271.
- SANDERSON, C. 2004. *Health Psychology*, John Wiley and Sons.
- SEXTON, D., BURRELL, B. & THOMPSON, B. 1992. Measurement Integrity of the Family Needs Survey. *Journal of Early Intervention*, 16, 343-352.
- SHARPE, D. & ROSSITER, L. 2002. Siblings of Children With a Chronic Illness: A Meta-Analysis. *Journal of Pediatric Psychology*, 27, 699-710.
- SHEERAN, T., MARVIN, R. S. & PIANTA, R. 1997. Mothers' Resolution of Their Child's Diagnosis and Self-Reported Measures of Parenting Stress, Marital Relations, and Social Support. *Journal of Pediatric Psychology*, 22, 197-212.
- SHINNAR, S., BERG, A. T., O'DELL, C., NEWSTEIN, D., MOSHE, S. L. & HAUSER, W. A. 2000. Predictors of multiple seizures in a cohort of children prospectively followed from the time of their first unprovoked seizure. *Annals of Neurology*, 48, 140-147.
- SHORE, C. P., AUSTIN, J. K., HUSTER, G. A. & DUNN, D. W. 2002. Identifying Risk Factors for Maternal Depression in Families of Adolescents With Epilepsy. *Journal for Specialists in Pediatric Nursing*, 7, 71-80.
- SIGN. 2003. Diagnosis and Management of Epilepsy in Adults. Available: <http://www.sign.ac.uk/pdf/sign70.pdf>.

- SIGN 2005. Diagnoses and management of epilepsies in children and young people. NHS Scotland.
- SMEDJE, H., BROMAN, J. E., HETTA, J. & VON KNORRING, A. L. 1999. Psychometric properties of a Swedish version of the "Strengths and Difficulties Questionnaire". *European Child & Adolescent Psychiatry*, 8, 63-70.
- SMITH, S. 1984. *Family Stress Theory: Review and Critique*. University of Georgia.
- SREEJA, I., GUPTA, S., RAKESH, L. & SINGH, M. B. 2009. Comparison Of Burden Between Family Caregivers Of Patients Having Schizophrenia And Epilepsy. *Internet Journal of Epidemiology*, 6, 2.
- STERLING, M. 2011. General Health Questionnaire - 28 (GHQ-28). *Journal of physiotherapy*, 57, 259.
- STEVANOVIC, D., JANCIC, J. & LAKIC, A. 2011. The impact of depression and anxiety disorder symptoms on the health-related quality of life of children and adolescents with epilepsy. *Epilepsia*, 52, e75-e78.
- STREISAND, R., BRANIECKI, S., TERCYAK, K. P. & KAZAK, A. E. 2001. Childhood illness-related parenting stress: The pediatric inventory for parents. *Journal of Pediatric Psychology*, 26, 155-162.
- STREISAND, R., KAZAK, A. E. & TERCYAK, K. P. 2003. Pediatric-Specific Parenting Stress and Family Functioning in Parents of Children Treated for Cancer. *Children's Health Care*, 32, 245-256.
- STREISAND, R., SWIFT, E., WICKMARK, T., CHEN, R. & HOLMES, C. S. 2005. Pediatric parenting stress among parents of children with type 1 diabetes: The role of self-efficacy, responsibility, and fear. *Journal of Pediatric Psychology*, 30, 513-521.
- THOMAS, S., BUTLER, R., HARE, D. J. & GREEN, D. 2011. Using personal construct theory to explore self-image with adolescents with learning disabilities. *British Journal of Learning Disabilities*, 39, 225-232.
- TRUTE, B. & HIEBERT-MURPHY, D. 2005. Predicting family adjustment and parenting stress in childhood disability services using brief assessment tools. *Journal of Intellectual and Developmental Disability*, 30, 217-225.
- TWENTE, U. O. 2010. *Transactional Model of Stress and Coping* [Online]. Available: http://www.utwente.nl/cw/theorieenoverzicht/Theory%20clusters/Health%20Communication/transactional_model_of_stress_and_coping.doc/ [Accessed 19/07/2011 2011].
- VARNI, J. W., BURWINKLE, T. M. & SEID, M. 2005. The PedsQL™ as a pediatric patient-reported outcome: Reliability and validity of the PedsQL™ measurement model in 25,000 children. *Expert Review of Pharmacoeconomics and Outcomes Research*, 5, 705-719.
- VARNI, J. W. & LIMBERS, C. A. 2009. The Pediatric Quality of Life Inventory: Measuring Pediatric Health-Related Quality of Life from the Perspective of Children and Their Parents. *Pediatric Clinics of North America*, 56, 843-863.
- WAGNER, J., SMITH, G., FERGUSON, P., BAKERGEM, K. & HRISKO, S. Pilot study of an integrated cognitive-behavioral and self-management intervention for youth with epilepsy and caregivers: Coping Openly and Personally with Epilepsy (COPE). *Epilepsy and Behavior*, 18, 280-285.
- WALKER, A. T. 1985. Reconceptualising Family Stress. *Journal of Marriage and Family*, 47, 827-837.
- WALLSTON, B. S., WALLSTON, K. A., KAPLAN, G. D. & MAIDES, S. A. 1976. Development and validation of the Health Locus of Control (HLC) Scale. *Journal of Consulting and Clinical Psychology*, 44, 580-585.
- WHO 2005. Atlas: Epilepsy care in the world. In: PROGRAMME FOR NEUROLOGICAL DISEASE AND NEUROSCIENCE, W. (ed.). Geneva: Global Campaign Against Epilepsy.
- WIEBE, S., CAMFIELD, P., JETTÉ, N. & BURNEO, J. G. 2009. Epidemiology of epilepsy: prevalence, impact, comorbidity and disparities. *The Canadian*

Journal Of Neurological Sciences. Le Journal Canadien Des Sciences Neurologiques, 36 Suppl 2, S7-S16.

WILLIAMS, J., STEEL, C., SHARP, G. B., DELOSREYES, E., PHILLIPS, T., BATES, S., LANGE, B. & GRIEBEL, M. L. 2003. Parental anxiety and quality of life in children with epilepsy. *Epilepsy & Behavior*, 4, 483-486.

Appendices:

Parent information sheet

Child <10 information sheet

Child >10 information sheet

Study questionnaires

PARTICIPANT INFORMATION SHEET

Parent

Correlates of stress adjustment in carers of children with newly diagnosed epilepsy.

We would like to invite you to take part in a 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.

(Part 1 tells you the purpose of the study and what will happen to you if you take part. Part 2 gives you more detailed information about the conduct of the study).

Part 1:

What is the purpose of the study?

Epilepsy is a common condition that affects 5 per 1000 children. It is increasingly recognised that even with effective treatment parents feel distressed by this diagnosis being given to their child. Non-medical interventions such as support workers may help with this distress but very little information exists to tell us how best to provide this sort of support.

The present study is designed to help us answer the question by looking at a number of different things that are known to increase parents' stress. These include family dynamics, parental coping strategies and support networks, symptoms of depression in the main carer, and the child's attitude towards themselves.

Why have I been invited to take part?

We have chosen you to take part in the study because you have been referred to a clinic in Alder Hey because a health professional feels that it is possible your child may have an epilepsy.

Do I have to take part?

It is up to you to decide. After you read this information leaflet and if you decide to participate in the study, we would ask you to contact one of the researchers via telephone (details below) to express your interest and ask any further questions you may have about the study. You are free to withdraw at any time, without having to give a reason.

What will happen to me if I take part?

The study is based at Alder Hey Children's Hospital and this is where the questionnaire meetings will take place.

If, after you have contacted the researcher to express your interest in the study, you would still like to take part, the researcher will contact you to arrange a meeting. At this meeting the researcher will go through this information sheet with you again and check that you still want to take part. If you do still wish to take part you will then be asked to sign a consent form to say you agree to taking part in the study. You will then be required to complete a set of questionnaires. This will take approximately 1 ½ hours. The management of your child's condition will proceed as normal. 5 months later another meeting will be arranged to complete the second set of questionnaires. These will be just the same as the first time and will also take approximately 1 ½ hours.

You will then have completed your involvement in the study.

What will I have to do?

The only thing you will have to do is be available with your child to complete the questionnaires at the time and place arranged with the researcher.

What are the possible disadvantages and risks of taking part?

There are no disadvantages or risks in taking part although it will take up a few hours of your time.

What are the possible benefits of taking part?

There are no direct benefits for you however, it is possible that completing the questionnaires may reveal health issues that can then be addressed.

Will taking part in the study be kept confidential?

Yes. We will follow ethical and legal practice and all information will be handled in confidence. The details are included in Part 2.

If the information in Part 1 has interested you and you are considering participation, please read the additional information in Part 2 before making any decision.

Part 2:

What if relevant new information becomes available?

It is possible that completing the questionnaires may reveal health issues that you were not aware of. If this should occur, then you will be fully informed of this and, with your permission, appropriate referrals will be made.

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

You are free to refuse to take part at any time, or to stop at any time, even during tests if these start to worry or tire you.

What if there is a problem?

Although very unlikely, if you are harmed by taking part in this research project, there are no special compensation arrangements. If you are harmed due to someone's negligence, then you may have grounds for a legal action but you

may have to pay for this. If you wish to make a complaint or have any concerns about any aspect of the way you have been approached or treated during the course of this study, please contact Dr Curran (details below) who will do his best to answer your questions. If you remain unhappy and wish to complain formally, you can do this via the normal complaints procedure. Details of which can be obtained from the hospital's website (www.alderhey.nhs.uk).

Will my taking part in this study be kept confidential?

All the information that is collected about you during the course of the research will be kept strictly confidential. If you agree to participate in this study, you are agreeing to personal data relating to yourself (as defined by the Data Protection Act, 1998), being used for research purposes. Your personal information will be kept and used solely by the researchers for up to ten years, and then will be confidentially destroyed. You have a legal right to view your personal information stored with us. If you wish to view your personal information, please write to Dr. Andrew Curran (see below for address) for more information on how to do this. If you decide that you no longer wish to take part in this study please inform Dr Andrew Curran on 07900 673645.

What will happen to the results of the research study?

We intend to publish the findings in relevant scientific journals and a copy can be made available to you at your request. You will not be personally identified in any publication.

Who is organising and funding the research?

The study is being run by Dr. Andrew Curran, consultant paediatric neurologist of the Royal Liverpool Children's Hospital.

Who has reviewed the study?

This study has been reviewed by the Royal Liverpool Children's Committee on Research Ethics and the Research Ethics Committee.

Who do I contact for further information?

If you require more information, please contact:
Dr Andrew Curran (Tel:07900 673645)
Department of Neurology,
Alder Hey Children's Hospital,
Eaton Road,
Liverpool
L12 2AP

To express an interest in the study, please contact either:
Dr Andrew Curran or Miss Louisa Bethell (MPhil student) – 0151 2525328

We would like to thank you for taking the time to read this information sheet.
Please do not hesitate to contact us if there are any questions that are unanswered.

PARTICIPANT INFORMATION SHEET

Children \leq 10 years

Stress in families of children with epilepsy

What is research? Why is the project being done?

Research is a way we try to find out the answers to questions.

Epilepsy is a word that describes why some people have seizures or funny turns. Lots of people in the world have funny turns, and lots of children do too. Having funny turns, especially if you have more than one, can be very upsetting for both you and your parents. We are doing this project because we want to see if we can help children and their families be less upset when they have these funny turns.

To do this we are going to look at a number of different things that we know can change whether children feel happy or upset. These include how you feel about things, how your family gets on together, how your mummy and daddy deal with stressful things, how well your mummy and daddy are and how much help they get from other people like your granny and grandpa.

Why have I been asked to take part?

We are asking you to take part in this study because your own doctor has asked for you to be seen in a clinic at Alder Hey Hospital because you have had at least one funny turn.

Do I have to take part?

If you want to take part you can. If you do not want to take part then you do not have to and nobody will be angry or upset with you. If you decide you do want to take part but then change your mind, that is ok and you will not have to do it anymore.

What will happen to me if I take part?

If you decide you want to take part, your parents can contact the people doing the study. We will then ask you and your parents to come in and meet us. We will ask you if you still want to do the study. If you still want to do the study we will ask you to answer some questions. The questions are not difficult, it's not like school! They are only questions about you and how you feel. This will take about 20 minutes. There will be someone there to help you.

The same person will also ask your parent to fill in some questionnaires which are about themselves and their feelings.

5 months after you have done this, we will want you to come and see us to answer the questions again. These will be just the same as the first time. Once again someone will help you to fill them in. They still only take 20 minutes to complete! You will then have finished your part in the study.

What will I have to do?

The only thing you will have to do is to fill in the questionnaires.

Might anything about the research upset me?

There is nothing in the research that will harm you. If you did get upset by any of the questions there will be someone there to talk to and you can stop at any time if you like.

Will joining in help me?

We cannot promise it will help you but the information we get might help other children who get funny turns in the future.

Will anyone else know I am doing this? Will my details be kept private?

Nobody will know that you are doing this study unless it is necessary that they do so. Nobody will know about the answers you have given in the questionnaires except the doctors doing the research. Your parent(s) would be told if we found something in your answers that we felt needed our help.

What will happen if I don't want to do it anymore?

If at any time you don't want to do the research anymore, just tell your parents, or the doctor. They will not be cross with you.

Who is organising the project?

The study is being run by Doctor Andrew Curran at Alder Hey Children's Hospital.

Did anyone else check the study is ok to do?

Before any research is allowed to happen, it has to be checked by a group of people called a Research Ethics Committee. This study has been checked by the Royal Liverpool Children's Committee on Research Ethics and the Research Ethics Committee.

Who do I talk to if I have any questions?

You can talk to your parents if you have any questions about the study and they can ask the doctor if there is anything they are not sure about.

Thank you for reading 😊

PARTICIPANT INFORMATION SHEET

Children > 10 years

Correlates of stress adjustment in carers of children with newly diagnosed epilepsy.

We would like to invite you to take part in a 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 your mother and father and anyone else about the study if you wish.

What is the purpose of the study?

Lots of young people have funny turns. In fact, the number of young people that have more than one funny turn is 5 per 1000. Having funny turns, especially if you have more than one can be very upsetting for both you and your parents. Professionals such as doctors and nurses can help young people and their families deal with the distress this causes but there is very little information to tell us how best to provide this sort of support.

The study we are asking you to take part in is designed to help us answer this question by looking at a number of different things that are known to increase the amount of distress young people and their parents feel. We know that there are a number of things that go on in a young person's life that can make distress better or worse. These include how you feel about things, how your family gets on together, how your mummy and daddy deal with stressful things in their own lives, how well your mummy and daddy are and how much help they get from other people like your granny and grandpa.

Why have I been invited to take part?

We are asking you to take part in this study because your own doctor has asked for you to be seen in a clinic at Alder Hey Hospital because you have had at least one funny turn.

Do I have to take part?

It is up to you to decide. After you read this information leaflet and if you decide to take part in the study, we would ask you or your parent to contact one of the researchers via telephone (details below) to express your interest and ask any further questions you may have about the study. You are free to stop being part of the study at any time, without having to give us a reason.

What will happen to me if I take part?

The study is based at Alder Hey Children's Hospital and this is where the questionnaire meetings will take place.

If, after you or your parent(s) have contacted the researcher to express your interest in the study, you would still like to take part, the researcher will contact your parent(s) to arrange a meeting with one of your parents and you. At this meeting the researcher will go through this information sheet with you again and check that you still want to take part. If you do still wish to take part you will then be asked to sign a consent form to say you agree to taking part in the study. You will then be required to fill out a short set of questionnaires. These are not difficult questionnaires. It's not like school! They are only to ask you some questions about yourself and how you are feeling. Filling out your questionnaires will take about 20 minutes. The person who comes out to visit will be there to help you. The same person will also ask your parent to fill in some questionnaires which are about themselves and their feelings. 5 months after you have done the first set of questionnaires, we will want you to complete a second set of questionnaires. These will be just the same as the first time. Once again the person who comes to see you and your parent will help you to fill them in. They still only take 20 minutes to complete! You will then have finished your part in the study.

What will I have to do?

The only thing you will have to do is to fill in the questionnaires.

What are the possible disadvantages and risks of taking part?

There are no disadvantages or risks in taking part although it will take up approximately an hour of your time.

What are the possible benefits of taking part?

There are no direct benefits for you however, it is possible that after you have filled in the questionnaires there will be some things about your feelings that you may want to discuss with someone. We can help you with that.

Will taking part in the study be kept confidential?

Yes. We will follow ethical and legal practice and all information will be handled in confidence. This means that nobody will know about the answers you have given in the questionnaires except the doctors doing the research. If you are under 16 years old, then your parent(s) would be told if we found something in your answers that we felt needed our help.

If you agree to participate in this study, you are agreeing to personal data relating to yourself (as defined by the Data Protection Act, 1998), being used for research purposes. Your personal information will be kept and used solely by the researchers for up to ten years, and then will be confidentially destroyed. You have a legal right to view your personal information stored with us. If you wish to view your personal information, please write to Dr. Andrew Curran (see below for address) for more information on how to do this. If you decide that you no longer wish to take part in this study please inform Dr Andrew Curran on 07900 673645.

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

You are free to refuse to take part at any time, or to stop at any time, even during tests if these start to worry or tire you.

What will happen to the results of the research study?

We intend to publish the findings in relevant scientific journals and a copy can be made available to you at your request. You will not be personally identified in any publication.

Who is organising and funding the research?

The study is being run by Dr. Andrew Curran, consultant paediatric neurologist of the Royal Liverpool Children's Hospital.

Who has reviewed the study?

This study has been reviewed by the Royal Liverpool Children's Committee on Research Ethics and the Research Ethics Committee.

Who do I contact for further information?

If you require more information, please contact:

Dr Andrew Curran (Tel:07900 673645)
Department of Neurology,
Alder Hey Children's Hospital,
Eaton Road,
Liverpool L12 2AP

To express an interest in the study, please contact either:

Dr Andrew Curran or Miss Louisa Bethell (MPhil student) – 0151 2525328

We would like to thank you for taking the time to read this information sheet.

Please do not hesitate to contact us if there are any questions that are unanswered.

Questionnaire Battery

'Fit' Event History

Date started:

How frequent are these events:

Characteristics:

When was the last event:

Have you had any investigations before this visit:

Is there any family history of epilepsy:

What do you (parent/carer) think is the problem:

PEDIATRIC INVENTORY FOR PARENTS

Below is a list of difficult events which parents of children who have (or have had) a serious illness sometimes face. Please read each event carefully, and circle HOW OFTEN the event has occurred for you in the past 7 days, using the 5 point scale below. Afterwards, please rate how DIFFICULT it was/or generally is for you, also using the 5 point scale. Please complete both columns for each item.

EVENT	HOW OFTEN?					HOW DIFFICULT?				
	1	2	3	4	5	1	2	3	4	5
1. Difficulty sleeping.....	1	2	3	4	5	1	2	3	4	5
2. Arguing with family member(s).....	1	2	3	4	5	1	2	3	4	5
3. Bringing my child to the clinic or hospital.....	1	2	3	4	5	1	2	3	4	5
4. Learning upsetting news	1	2	3	4	5	1	2	3	4	5
5. Being unable to go to work/job	1	2	3	4	5	1	2	3	4	5
6. Seeing my child's mood change quickly.....	1	2	3	4	5	1	2	3	4	5
7. Speaking with doctor.....	1	2	3	4	5	1	2	3	4	5
8. Watching my child have trouble eating.....	1	2	3	4	5	1	2	3	4	5
9. Waiting for my child's test results	1	2	3	4	5	1	2	3	4	5
10. Having money/financial troubles	1	2	3	4	5	1	2	3	4	5
11. Trying not to think about my family's difficulties.....	1	2	3	4	5	1	2	3	4	5
12. Feeling confused about medical information	1	2	3	4	5	1	2	3	4	5
13. Being with my child during medical procedures.....	1	2	3	4	5	1	2	3	4	5
14. Knowing my child is hurting or in pain	1	2	3	4	5	1	2	3	4	5
15. Trying to attend to the needs of other family members.....	1	2	3	4	5	1	2	3	4	5
16. Seeing my child sad or scared	1	2	3	4	5	1	2	3	4	5
17. Talking with the nurse	1	2	3	4	5	1	2	3	4	5
18. Making decisions about medical care or medicines	1	2	3	4	5	1	2	3	4	5
19. Thinking about my child being isolated from others.....	1	2	3	4	5	1	2	3	4	5
20. Being far away from family and/or friends	1	2	3	4	5	1	2	3	4	5
21. Feeling numb inside.....	1	2	3	4	5	1	2	3	4	5
22. Disagreeing with a member of the health care team	1	2	3	4	5	1	2	3	4	5
23. Helping my child with his/her hygiene needs.....	1	2	3	4	5	1	2	3	4	5
24. Worrying about the long term impact of the illness.....	1	2	3	4	5	1	2	3	4	5
25. Having little time to take care of my own needs	1	2	3	4	5	1	2	3	4	5

EVENT	HOW OFTEN?					HOW DIFFICULT?				
	1=Never, 2=Rarely, 3=Sometimes, 4=Often, 5=Very often					1=Not at all, 2=A little, 3=Somewhat, 4=Very much, 5=Extremely				
26. Feeling helpless over my child's condition	1	2	3	4	5	1	2	3	4	5
27. Feeling misunderstood by family/friends as to the severity of my child's illness	1	2	3	4	5	1	2	3	4	5
28. Handling changes in my child's daily medical routines	1	2	3	4	5	1	2	3	4	5
29. Feeling uncertain about the future.....	1	2	3	4	5	1	2	3	4	5
30. Being in the hospital over weekends/holidays	1	2	3	4	5	1	2	3	4	5
31. Thinking about other children who have been seriously ill.....	1	2	3	4	5	1	2	3	4	5
32. Speaking with child about his/her illness	1	2	3	4	5	1	2	3	4	5
33. Helping my child with medical procedures (e.g. giving shots, swallowing medicine, changing dressing)	1	2	3	4	5	1	2	3	4	5
34. Having my heart beat fast, sweating, or feeling tingly.....	1	2	3	4	5	1	2	3	4	5
35. Feeling uncertain about disciplining my child	1	2	3	4	5	1	2	3	4	5
36. Feeling scared that my child could get very sick or die.....	1	2	3	4	5	1	2	3	4	5
37. Speaking with family members about my child's illness	1	2	3	4	5	1	2	3	4	5
38. Watching my child during procedures	1	2	3	4	5	1	2	3	4	5
39. Missing important events in the lives of other family members.	1	2	3	4	5	1	2	3	4	5
40. Worrying about how friends and relatives interact with my child	1	2	3	4	5	1	2	3	4	5
41. Noticing a change in my relationship with my partner.....	1	2	3	4	5	1	2	3	4	5
42. Spending a great deal of time in unfamiliar settings	1	2	3	4	5	1	2	3	4	5

Family Questionnaire No. 1

Thank you for agreeing to complete a questionnaire about your child and family for a research project. We are grateful to you for giving us your time. The purpose of the questionnaire is to help us have a better understanding of your child's problems and how this is affecting him/her and also the rest of the family. We understand how stressful it can be to have a child with epilepsy and hope that the identification of specific difficulties faced by families with such children will help us to improve the care we provide and in turn, the quality of life for the whole family, not just the child.

If you have concerns regarding any of the questions or need help to complete the questionnaire please contact one of our researchers who will be pleased to assist.

Please tick/circle and give details where applicable. Here is a list of different levels of stress for you to refer to in answering these questions:

0=None 1=A bit 2=Somewhat 3=Fairly 4=A lot

Child's Name:

Your Name:

Relationship to child:

Name and age of other family members who live in your household:

.....
.....
.....

Are any other family members e.g. grandparents, or friends closely involved in the care of your child? Please give their name(s) and relationship to the child:

.....
.....
.....
.....

1. During the past 12 months have you considered alternative therapies for your child? (Please circle) YES/NO

Did you take any of them up? YES/NO

If yes, what have you tried?
.....

2. Does your child have any physical health problems? (Please circle) YES/NO
(movement, hearing, vision, asthma, allergies)

If yes, describe

3. Does your child have any behaviour problems? YES/NO
(hyperactive, concentration, aggression)

If yes, describe

4. Does your child have any cognitive problems? YES/NO
(language delay, learning difficulty)

If yes, describe

5. Does your child have any problems achieving at school compared with the majority of children of the same age? YES/NO

If yes, describe
.....
.....

6. Does your child have a diagnosis for his/her condition? (Please circle) YES/NO

If yes, what diagnosis does your child have?
.....

If no, would you like to add a comment?

.....
.....
.....
.....
.....

7. Is it difficult for you to manage the problems your child has? YES/NO

How much stress has this caused you? 0 1 2 3 4

8. Have there been any difficulties with the school/nursery meeting your child's needs in regard to these problems? YES/NO NOT APPLICABLE

How stressful is this for you? 0 1 2 3 4

Comments.....
...
.....
...

9. During the past 12 months have you seen any of the following? (Please circle)

Paediatrician	<u>YES/NO</u>	How many times? Appointment length?..... mins
GP	<u>YES/NO</u>	How many times? Appointment length?..... mins
Physiotherapist	<u>YES/NO</u>	How many times? Appointment length?..... mins
Occupational therapist	<u>YES/NO</u>	How many times? Appointment length?..... mins
Speech and language therapist	<u>YES/NO</u>	How many times? Appointment length?..... mins
Social worker	<u>YES/NO</u>	How many times? Appointment length?..... mins
Health visitor	<u>YES/NO</u>	How many times? Appointment length?..... mins
Other (specify).....	<u>YES/NO</u>	How many times? Appointment length?..... mins

Are you currently on a waiting list for any of the above? YES/NO

If yes please specify.

.....
.....
.....

10. During the past 12 months how many GP/hospital/education appointments have you attended with your child in total?

How stressful has this been for you? (Please circle) 0 1 2 3 4

Comments.....
...
.....
...

I 1. If in paid employment have you been granted leave in the past 12 months to attend appointments concerning your child? (Please circle) YES/NO

How stressful has this been for you? 0 1 2 3 4

Comments.....
...

I3. What type of leave is normally granted by your employer? (Please tick)

paid() unpaid() compassionate() dependency() holiday()
not applicable()

Comments.....
...
.....
...

I4. Has your child's condition had an effect on your ability to carry out paid work during the past 12 months? (Please circle) YES/NO NOT APPLICABLE

How stressful has this been for you? 0 1 2 3 4

Comments.....
...
.....
...

I5. Have you had to incur travel costs to attend appointments? YES/NO

How stressful has this been for you? 0 1 2 3 4

Comments.....
...
.....
...

Please estimate how much you have spent in the last 12 months.....

16. Has your child's condition had an effect upon your general financial situation during the past 12 months? YES/NO

How stressful has this been for you? 0 1 2 3 4

Comments.....
...
.....
...

17. Has your child's condition affected your social life in the past 12 months?

YES/NO

How stressful has this been for you? 0 1 2 3 4

Comments.....
...
.....
...

18. Has your spouse/partner had any difficulties accepting or dealing with the problems arising from your child's condition?

YES/NO

How stressful has this been for you? 0 1 2 3 4

Comments.....
...

19. Have these problems made it difficult for you to find time to spend with your spouse/partner? YES/NO

How stressful has this been for you? 0 1 2 3 4

Comments.....
...
.....
...

20. How do you feel about your child's condition now? (Please tick)

Optimistic () Pessimistic() Don't know()

How stressful is this for you? 0 1 2 3 4

Comments.....
...

.....
...

21. Has your child's condition affected his/her acceptance by peers? YES/NO

How stressful has this been for you? 0 1 2 3 4

Comments.....
...
.....
...

22. Have the problems your child experiences affected how he/she gets along with other children? *(Please circle)*
YES/NO

How stressful is this for you? 0 1 2 3 4

Comments.....
...
.....
...

23. Has your child's condition resulted in he/she being more dependent on you, or needing more supervision, than other children of the same age?
YES/NO

How stressful is this for you? 0 1 2 3 4

Comments.....
...
.....
...

24. Do you believe your child's self esteem is lower because of his/her problems?

How stressful is this for you? 0 1 2 3 4 YES/NO

Comments.....
...
.....
...

25. Are you concerned that this problem will affect your child's future?
YES/NO

How stressful is this for you?

0 | 2 3 4

Comments.....

...

.....

...

26. Have sibling(s) had any difficulties accepting or dealing with these problems?

YES/NO

How stressful has this been for you?

0 | 2 3 4

Comments.....

...

.....

...

27. If you have any other children, has this problem made it difficult for you to spend time with them?

YES/NO

How stressful has this been for you?

0 | 2 3 4

Comments.....

...

.....

...

28. Has this problem made it difficult for the family to spend time together?

YES/NO

How stressful has this been for you?

0 | 2 3 4

Comments.....

...

.....

...

29. Due to your child's condition do you find any of the following activities difficult? (Please circle)

Shopping

YES/NO

If yes, how stressful is this? 0 | 2 3 4

Visiting family and friends

YES/NO

If yes, how stressful is this? 0 | 2 3 4

Swimming

YES/NO

If yes, how stressful is this? 0 | 2 3 4

Parks and playgrounds

YES/NO

If yes, how stressful is this? 0 | 2 3 4

Church	<u>YES/NO</u>	If yes, how stressful is this? 0 1 2 3
4		
Other.....	<u>YES/NO</u>	If yes, how stressful is this? 0 1
2 3 4		

Comments.....

...

.....

...

30. What would you *most* like to do as a family that is currently difficult due to your child's condition?

.....

....

.....

....

Thank you for your time and effort in filling out this questionnaire.

THE GENERAL HEALTH QUESTIONNAIRE

GHQ 28

David Goldberg

Please read this carefully.

We should like to know if you have had any medical complaints and how your health has been in general, over the past few weeks. Please answer ALL the questions on the following pages simply by underlining the answer which you think most nearly applies to you. Remember that we want to know about present and recent complaints, not those that you had in the past.

It is important that you try to answer ALL the questions.

Thank you very much for your co-operation.

Have you recently

A1 – been feeling perfectly well and in good health?	Better than usual	Same as usual	Worse than usual	Much worse than usual
A2 – been feeling in need of a good tonic?	Not at all	No more than usual	Rather more than usual	Much more than usual
A3 – been feeling run down and out of sorts?	Not at all	No more than usual	Rather more than usual	Much more than usual
A4 – felt that you are ill?	Not at all	No more than usual	Rather more than usual	Much more than usual
A5 – been getting any pains in your head?	Not at all	No more than usual	Rather more than usual	Much more than usual
A6 – been getting a feeling of tightness or pressure in your head?	Not at all	No more than usual	Rather more than usual	Much more than usual
A7 – been having hot or cold spells?	Not at all	No more than usual	Rather more than usual	Much more than usual
B1 – lost much sleep over worry?	Not at all	No more than usual	Rather more than usual	Much more than usual
B2 – had difficulty in staying asleep once you are off?	Not at all	No more than usual	Rather more than usual	Much more than usual
B3 – felt constantly under strain?	Not at all	No more than usual	Rather more than usual	Much more than usual
B4 – been getting edgy and bad-tempered?	Not at all	No more than usual	Rather more than usual	Much more than usual
B5 – been getting scared or panicky for no good reason?	Not at all	No more than usual	Rather more than usual	Much more than usual
B6 – found everything getting on top of you?	Not at all	No more than usual	Rather more than usual	Much more than usual
B7 – been feeling nervous and strung-up all the time?	Not at all	No more than usual	Rather more than usual	Much more than usual

Please turn over



Have you recently

C1 – been managing to keep yourself busy and occupied?	More so than usual	Same as usual	Rather less than usual	Much less than usual
C2 – been taking longer over the things you do?	Quicker than usual	Same as usual	Longer than usual	Much longer than usual
C3 – felt on the whole you were doing things well?	Better than usual	About the same	Less well than usual	Much less well
C4 – been satisfied with the way you've carried out your task?	More satisfied	About same as usual	Less satisfied than usual	Much less satisfied
C5 – felt that you are playing a useful part in things?	More so than usual	Same as usual	Less useful than usual	Much less useful
C6 – felt capable of making decisions about things?	More so than usual	Same as usual	Less so than usual	Much less capable
C7 – been able to enjoy your normal day-to-day activities?	More so than usual	Same as usual	Less so than usual	Much less than usual

D1 – been thinking of yourself as a worthless person?	Not at all	No more than usual	Rather more than usual	Much more than usual
D2 – felt that life is entirely hopeless?	Not at all	No more than usual	Rather more than usual	Much more than usual
D3 – felt that life isn't worth living?	Not at all	No more than usual	Rather more than usual	Much more than usual
D4 – thought of the possibility that you might make away with yourself?	Definitely not	I don't think so	Has crossed my mind	Definitely have
D5 – found at times you couldn't do anything because your nerves were too bad?	Not at all	No more than usual	Rather more than usual	Much more than usual
D6 – found yourself wishing you were dead and away from it all?	Not at all	No more than usual	Rather more than usual	Much more than usual
D7 – found that the idea of taking your own life kept coming into your mind?	Definitely not	I don't think so	Has crossed my mind	Definitely has

A
 B
 C
 D
 TOTAL

Rotter's Locus of Control Scale

1. a. Children get into trouble because their parents punish them too much.
1. b. The trouble with most children nowadays is that their parents are too easy with them.
2. a. Many of the unhappy things in people's lives are partly due to bad luck.
2. b. People's misfortunes result from the mistakes they make.
3. a. One of the major reasons why we have wars is because people don't take enough interest in politics.
3. b. There will always be wars, no matter how hard people try to prevent them.
4. a. In the long run people get the respect they deserve in this world.
4. b. Unfortunately, an individual's worth often passes unrecognized no matter how hard he tries.
5. a. The idea that teachers are unfair to students is nonsense.
5. b. Most students don't realize the extent to which their grades are influenced by accidental happenings.
6. a. Without the right breaks, one cannot be an effective leader.
6. b. Capable people who fail to become leaders have not taken advantage of their opportunities.
7. a. No matter how hard you try, some people just don't like you.
7. b. People who can't get others to like them don't understand how to get along with others.
8. a. Heredity plays the major role in determining one's personality.
8. b. It is one's experiences in life which determine what they're like.
9. a. I have often found that what is going to happen will happen.
9. b. Trusting fate has never turned out as well for me as making a decision to take a definite course of action.
10. a. In the case of the well prepared student there is rarely, if ever, such a thing as an unfair test.
10. b. Many times, exam questions tend to be so unrelated to course work that studying is really useless.
11. a. Becoming a success is a matter of hard work, luck has little or nothing to do with it.
11. b. Getting a good job depends mainly on being in the right place at the right time.
12. a. The average citizen can have an influence in government decisions.
12. b. This world is run by the few people in power, and there is not much the little guy can do about it.
13. a. When I make plans, I am almost certain that I can make them work.
13. b. It is not always wise to plan too far ahead because many things turn out to be a matter of good or bad fortune anyhow. 1

- 14. a. There are certain people who are just no good.
- 14. b. There is some good in everybody.
- 15. a. In my case getting what I want has little or nothing to do with luck.
- 15. b. Many times we might just as well decide what to do by flipping a coin.
- 16. a. Who gets to be the boss often depends on who was lucky enough to be in the right place first.
- 16. b. Getting people to do the right thing depends upon ability - luck has little or nothing to do with it.
- 17. a. As far as world affairs are concerned, most of us are the victims of forces we can neither understand, nor control.
- 17. b. By taking an active part in political and social affairs the people can control world events.
- 18. a. Most people don't realize the extent to which their lives are controlled by accidental happenings.
- 18. b. There really is no such thing as "luck."
- 19. a. One should always be willing to admit mistakes.
- 19. b. It is usually best to cover up one's mistakes.
- 20. a. It is hard to know whether or not a person really likes you.
- 20. b. How many friends you have depends upon how nice a person you are.
- 21. a. In the long run the bad things that happen to us are balanced by the good ones.
- 21. b. Most misfortunes are the result of lack of ability, ignorance, laziness, or all three.
- 22. a. With enough effort we can wipe out political corruption.
- 22. b. It is difficult for people to have much control over the things politicians do in office.
- 23. a. Sometimes I can't understand how teachers arrive at the grades they give.
- 23. b. There is a direct connection between how hard I study and the grades I get.
- 24. a. A good leader expects people to decide for themselves what they should do.
- 24. b. A good leader makes it clear to everybody what their jobs are.
- 25. a. Many times I feel that I have little influence over the things that happen to me.
- 25. b. It is impossible for me to believe that chance or luck plays an important role in my life.
- 26. a. People are lonely because they don't try to be friendly.
- 26. b. There's not much use in trying too hard to please people, if they like you, they like you.
- 27. a. There is too much emphasis on athletics in high school.
- 27. b. Team sports are an excellent way to build character.
- 28. a. What happens to me is my own doing.

28. b. Sometimes I feel that I don't have enough control over the direction my life is taking.

29. a. Most of the time I can't understand why politicians behave the way they do.

29. b. In the long run the people are responsible for bad government on a national as well as on a local level.

Score one point for each of the following:

2.a, 3.b, 4.b, 5.b, 6.a, 7.a, 9.a, 10.b, 11.b, 12.b, 13.b, 15.b, 16.a, 17.a, 18.a, 20.a, 21.a, 22.b, 23.a, 25.a, 26.b, 28.b, 29.a.

A high score = External Locus of Control

A low score = Internal Locus of Control

Locus of Control

Locus of Control refers to the extent to which individuals believe that they can control events that affect them. Individuals with a high *internal locus of control* believe that events result primarily from their own behaviour and actions. Those with a high *external locus of control* believe that powerful others, fate, or chance primarily determine events. Those with a high *internal locus of control* have better control of their behaviour and tend to exhibit more political behaviours than *externals* and are more likely to attempt to influence other people; they are more likely to assume that their efforts will be successful. They are more active in seeking information and knowledge concerning their situation than do *externals*. The propensity to engage in political behaviour is stronger for individuals who have a high *internal locus of control* than for those who have a high *external locus of control*.

Brief COPE

These items deal with ways you've been coping with the stress in your life since you found out you were going to have to have this operation. There are many ways to try to deal with problems. These items ask what you've been doing to cope with this one. Obviously, different people deal with things in different ways, but I'm interested in how you've tried to deal with it. Each item says something about a particular way of coping. I want to know to what extent you've been doing what the item says. How much or how frequently. Don't answer on the basis of whether it seems to be working or not—just whether or not you're doing it. Use these response choices. Try to rate each item separately in your mind from the others. Make your answers as true FOR YOU as you can.

- 1 = I haven't been doing this at all
- 2 = I've been doing this a little bit
- 3 = I've been doing this a medium amount
- 4 = I've been doing this a lot

1. I've been turning to work or other activities to take my mind off things.
2. I've been concentrating my efforts on doing something about the situation I'm in.
3. I've been saying to myself "this isn't real."
4. I've been using alcohol or other drugs to make myself feel better.
5. I've been getting emotional support from others.
6. I've been giving up trying to deal with it.
7. I've been taking action to try to make the situation better.
8. I've been refusing to believe that it has happened.
9. I've been saying things to let my unpleasant feelings escape.
10. I've been getting help and advice from other people.
11. I've been using alcohol or other drugs to help me get through it.
12. I've been trying to see it in a different light, to make it seem more positive.
13. I've been criticizing myself.
14. I've been trying to come up with a strategy about what to do.
15. I've been getting comfort and understanding from someone.
16. I've been giving up the attempt to cope.
17. I've been looking for something good in what is happening.
18. I've been making jokes about it.
19. I've been doing something to think about it less, such as going to movies, watching TV, reading, daydreaming, sleeping, or shopping.
20. I've been accepting the reality of the fact that it has happened.
21. I've been expressing my negative feelings.
22. I've been trying to find comfort in my religion or spiritual beliefs.
23. I've been trying to get advice or help from other people about what to do.
24. I've been learning to live with it.
25. I've been thinking hard about what steps to take.
26. I've been blaming myself for things that happened.
27. I've been praying or meditating.
28. I've been making fun of the situation.

	Not available	Not helpful at all	Sometimes helpful	Generally helpful	Very helpful	Extremely helpful
My parents						

Family Support Scale

Who gives you support and how would you rate it? Put a "Y" in the corresponding box.

My partner's parents						
My relatives						
My partner's relatives						
Partner						
My friends						
My partner's friends						
My children						
Other parents						
Co-workers						
Parent groups						
Social groups						
Places of worship						
My family or child's doctor						
Professional helpers (social workers, therapists, teachers, etc.)						
Professional agencies (public health, social services, mental health etc)						
School/ day-care centre						
Early intervention programme						

Family Needs Survey

(Revised, 1990b)

Child's Name: _____ Person Completing Survey: _____

Date Completed: ____/____/____ Relationship to Child: _____

Dear Parent:

Many families of young children have needs for information or support. If you wish, our staff are very willing to

discuss these needs with you and work with you to identify resources that might be helpful.

Listed below are some needs commonly expressed by families. It would be helpful to us if you would check

in the columns on the right any topics you would like to discuss. At the end there is a place for you to describe

other topics not included in the list.

If you choose to complete this form, the information you provide will be kept confidential. If you would prefer

not to complete the survey at this time, you may keep it for your records.

Would you like to discuss this topic
with a staff person from our program?

TOPICS No

Not

Sure Yes

Information

1. How children grow and develop
2. How to play or talk with my child
3. How to teach my child
4. How to handle my child's behavior
5. Information about any condition or disability my child might have
6. Information about services that are presently available for my child
7. Information about the services my child might receive in the future

Family & Social Support

1. Talking with someone in my family about concerns
2. Having friends to talk to
3. Finding more time for myself
4. Helping my spouse accept any condition our child might have
5. Helping our family discuss problems and reach solutions
6. Helping our family support each other during difficult times
7. Deciding who will do household chores, child care, and other family tasks
8. Deciding on and doing family recreational activities

Financial

1. Paying for expenses such as food, housing, medical care, clothing, or transportation
2. Getting any special equipment my child needs
3. Paying for therapy, day care, or other services my child needs
4. Counseling or help in getting a job
5. Paying for babysitting or respite care

Would you like to discuss this topic
with a staff person from our program?

TOPICS No

Not

Sure Yes

Explaining to Others

1. Explaining my child's condition to my parents or my spouse's parents
2. Explaining my child's condition to his or her siblings
3. Knowing how to respond when friends, neighbors, or strangers ask questions about my child
4. Explaining my child's condition to other children
5. Finding reading material about other families who have a child like mine

Child Care

1. Locating babysitters or respite care providers who are willing and able to care for my child.
2. Locating a day care program or preschool for my child
3. Getting appropriate care for my child in a church or synagogue during religious services

Professional Support

1. Meeting with a minister, priest, or rabbi
2. Meeting with a counselor (psychologist, social worker, psychiatrist)
3. More time to talk to my child's teacher or therapist

Community Services

1. Meeting & talking with other parents who have a child like mine
2. Locating a doctor who understands me and my child's needs
3. Locating a dentist who will see my child

Other: Please list other topics or provide any other information that you would like to discuss .

Is there a particular person with whom you would prefer to meet?

Thank you for your time.

We hope this form will be helpful to you in identifying the services that you feel are important.

FACES IV: Questionnaire

Directions to Family Members:

1. All family members over the age 12 can complete FACES IV.
2. Family members should complete the instrument independently, not consulting or discussing their responses until they have been completed.
3. Fill in the corresponding **number** in the space on the provided answer sheet.

1	2	3	4	5
Strongly Disagree	Generally Disagree	Undecided Generally	Agree	Strongly Agree

1. Family members are involved in each others lives.
2. Our family tries new ways of dealing with problems.
3. We get along better with people outside our family than inside.
4. We spend too much time together.
5. There are strict consequences for breaking the rules in our family.
6. We never seem to get organized in our family.
7. Family members feel very close to each other.
8. Parents equally share leadership in our family.
9. Family members seem to avoid contact with each other when at home.
10. Family members feel pressured to spend most free time together.
11. There are clear consequences when a family member does something wrong.
12. It is hard to know who the leader is in our family.
13. Family members are supportive of each other during difficult times.
14. Discipline is fair in our family.
15. Family members know very little about the friends of other family members.
16. Family members are too dependent on each other.
17. Our family has a rule for almost every possible situation.
18. Things do not get done in our family.
19. Family members consult other family members on important decisions.
20. My family is able to adjust to change when necessary.
21. Family members are on their own when there is a problem to be solved.
22. Family members have little need for friends outside the family.
23. Our family is highly organized.
24. It is unclear who is responsible for things (chores, activities) in our family.
25. Family members like to spend some of their free time with each other.
26. We shift household responsibilities from person to person.
27. Our family seldom does things together.
28. We feel too connected to each other.
29. Our family becomes frustrated when there is a change in our plans or routines.
30. There is no leadership in our family.
31. Although family members have individual interests, they still participant in family activities.
32. We have clear rules and roles in our family.
33. Family members seldom depend on each other.
34. We resent family members doing things outside the family.
35. It is important to follow the rules in our family.

36. Our family has a hard time keeping track of who does various household tasks.
37. Our family has a good balance of separateness and closeness.
38. When problems arise, we compromise.
39. Family members mainly operate independently.
40. Family members feel guilty if they want to spend time away from the family.
41. Once a decision is made, it is very difficult to modify that decision.
42. Our family feels hectic and disorganized.
43. Family members are satisfied with how they communicate with each other.
44. Family members are very good listeners.
45. Family members express affection to each other.
46. Family members are able to ask each other for what they want.
47. Family members can calmly discuss problems with each other.
48. Family members discuss their ideas and beliefs with each other.
49. When family members ask questions of each other, they get honest answers.
50. Family members try to understand each other's feelings
51. When angry, family members seldom say negative things about each other.
52. Family members express their true feelings to each other.

1	2	3	4	5
Very dissatisfied	Somewhat dissatisfied	Generally satisfied	Very satisfied	Extremely satisfied

How satisfied are you with:

53. The degree of closeness between family members.
54. Your family's ability to cope with stress.
55. Your family's ability to be flexible.
56. Your family's ability to share positive experiences.
57. The quality of communication between family members.
58. Your family's ability to resolve conflicts.
59. The amount of time you spend together as a family.
60. The way problems are discussed.
61. The fairness of criticism in your family.
62. Family members concern for each other.

Thank you for Your Cooperation!

Strengths and Difficulties Questionnaire

P 4-16

For each item, please mark the box for Not True, Somewhat True or Certainly True. It would help us if you answered all items as best you can even if you are not absolutely certain or the item seems daft! Please give your answers on the basis of the child's behaviour over the last six months.

Child's Name

Male/Female

Date of Birth.....

	Not True	Somewhat True	Certainly True
Considerate of other people's feelings	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Restless, overactive, cannot stay still for long	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often complains of headaches, stomach-aches or sickness	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Shares readily with other children (treats, toys, pencils etc.)	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often has temper tantrums or hot tempers	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Rather solitary, tends to play alone	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Generally obedient, usually does what adults request	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Many worries, often seems worried	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Helpful if someone is hurt, upset or feeling ill	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Constantly fidgeting or squirming	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Has at least one good friend	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often fights with other children or bullies them	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often unhappy, down-hearted or tearful	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Generally liked by other children	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Easily distracted, concentration wanders	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Nervous or clingy in new situations, easily loses confidence	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Kind to younger children	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often lies or cheats	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Picked on or bullied by other children	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often volunteers to help others (parents, teachers, other children)	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Thinks things out before acting	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Steals from home, school or elsewhere	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Gets on better with adults than with other children	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Many fears, easily scared	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Sees tasks through to the end, good attention span	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

Do you have any other comments or concerns?

Overall, do you think that your child has difficulties in one or more of the following areas:
emotions, concentration, behaviour or being able to get on with other people?

No	Yes- minor difficulties	Yes- definite difficulties	Yes- severe difficulties
<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

If you have answered "Yes", please answer the following questions about these difficulties:

- How long have these difficulties been present?

Less than a month	1-5 months	6-12 months	Over a year
<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

- Do the difficulties upset or distress your child?

Not at all	Only a little	Quite a lot	A great deal
<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

- Do the difficulties interfere with your child's everyday life in the following areas?

	Not at all	Only a little	Quite a lot	A great deal
HOME LIFE	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
FRIENDSHIPS	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
CLASSROOM LEARNING	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
LEISURE ACTIVITIES	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

- Do the difficulties put a burden on you or the family as a whole?

Not at all	Only a little	Quite a lot	A great deal
<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

Signature

Date

In the past **ONE month**, how much of a **problem** has this been for you ...

ABOUT MY HEALTH AND ACTIVITIES (problems with...)	Never	Almost Never	Some- times	Often	Almost Always
1. It is hard for me to walk more than one block	0	1	2	3	4
2. It is hard for me to run	0	1	2	3	4
3. It is hard for me to do sports activity or exercise	0	1	2	3	4
4. It is hard for me to lift something heavy	0	1	2	3	4
5. It is hard for me to take a bath or shower by myself	0	1	2	3	4
6. It is hard for me to do chores around the house	0	1	2	3	4
7. I hurt or ache	0	1	2	3	4
8. I have low energy	0	1	2	3	4

ABOUT MY FEELINGS (problems with...)	Never	Almost Never	Some- times	Often	Almost Always
1. I feel afraid or scared	0	1	2	3	4
2. I feel sad or blue	0	1	2	3	4
3. I feel angry	0	1	2	3	4
4. I have trouble sleeping	0	1	2	3	4
5. I worry about what will happen to me	0	1	2	3	4

HOW I GET ALONG WITH OTHERS (problems with...)	Never	Almost Never	Some- times	Often	Almost Always
1. I have trouble getting along with other kids	0	1	2	3	4
2. Other kids do not want to be my friend	0	1	2	3	4
3. Other kids tease me	0	1	2	3	4
4. I cannot do things that other kids my age can do	0	1	2	3	4
5. It is hard to keep up when I play with other kids	0	1	2	3	4

ABOUT SCHOOL (problems with...)	Never	Almost Never	Some- times	Often	Almost Always
1. It is hard to pay attention in class	0	1	2	3	4
2. I forget things	0	1	2	3	4
3. I have trouble keeping up with my schoolwork	0	1	2	3	4
4. I miss school because of not feeling well	0	1	2	3	4
5. I miss school to go to the doctor or hospital	0	1	2	3	4

