Health Related Stress Measured In Carers Of Children With Possible Epilepsy:
Interim Analysis Of The Stress Adjustment In Carers Of Children Who Are Newly Diagnosed With Epilepsy

"Stress And Its Covariates In Parents Who Have Been Referred To Clinic With A Possible Diagnosis Of Epilepsy In Their Child"

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Abstract

Background: Parenting Stress has been found to be associated with children who have been diagnosed with epilepsy. However the percentage of parents feeling stressed differs across studies that have examined this relationship.

Aims: To examine carer stress when their child has been referred to Alder Hey Children's Hospital with epilepsy being a suggested diagnosis. The study then aims to examine for associations between different factors and carer stress.

Methods: To recruit carers of children aged 0 to 16 years of age, who have a queried diagnosis of epilepsy and have been referred to an outpatient department at Alder Hey Children's Hospital. A battery of 9 questionnaires will be given to the carer along with a demographic screen. The questionnaire battery will measure carer health-related stress and factors that may or may not affect whether stress is being felt. The factors being examined are carer's recent health, how they cope with the potential diagnosis, the degree of control they feel they have over events, the amount of support they receive and would like to get, their family environment and their opinion of their child's behaviour.
Two questionnaires will be given to the child to measure their self-esteem, self-image and their quality of life. If they are aged 6 years only one can be completed but if aged over 6 years both questionnaires can be completed.
A statistical analysis will be performed on the questionnaires completed by the carer. A Spearman's Rank correlation will examine associations between the stress score and the scores of the other questionnaires. A Mann Whitney U test will assess for differences between questionnaire scores in those classified as either stressed or not stressed and a binary logistic regression will be used to form a model of questionnaires likely to predict higher carer stress.

Results: 133 carers were approached with 60 carers giving consent, however 6 carers withdrew from the study resulting in 54 carers recruited during this study year. Results from the previous year's recruitment were included in the analysis, bringing the total number of carers involved in the study to be 59.
Both Stress questionnaires revealed high stress scores being reported by the carers. The Pediatric Inventory for Parents (PIP) categorised 43% of carers as being stressed, with the semi-structured stress questionnaire categorising 75% of parents being stressed. A binary logistic regression model revealed two predictive models by finding that the Strengths and Difficulties Questionnaire (SDQ) along with either the Family Needs Survey (FNS) or the Brief COPE Inventory subgroup dysfunctional coping strategy was able to predict high PIP stress scores.

Conclusion: This study revealed that a high percentage of carers were experiencing stress. The binary logistic regression model suggests that the questionnaires most useful in predicting high carer stress scores were the SDQ with either the FNS or the Brief COPE Inventory. This guides future interventions as by using these questionnaires along with the PIP, carers would benefit from extra assistance can be targeted in order to prevent stress becoming problematic.

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List of Abbreviations

AED	Anti-Epileptic Drug
BBFM	Biobehavioural Family Model
CIDI	Composite International Diagnostic Instrument
COPE	Coping Orientations to Problems Experienced
CWE	Children With Epilepsy
EEG	Electroencephalograph
FAAR	Family Adjustment and Adaptability Response
FACES	Family Adjustment and Cohesive Evaluation Scale
FBII	Family Burden of Injury interview
FILE	Family Inventory of Life Events and Changes
FNS	Family Needs Survey
FSS	Family Support Scale
GAS	General Adaptation Syndrome
GHQ	General Health Questionnaire
HRQOL	Health Related Quality of Life
ILAE	International league Against Epilepsy
LEDS	Life Events and Difficulty Schedule
MRI	Magnetic Resonance Imaging
NGPSE	National general Practice Study of Epilepsy
NICE	National Institute of Clinical Excellence
PedsQL	Pediatric Quality of Life Inventory
PIP	Pediatric Inventory for Parents
PSI	Parenting Stress Index
SD	Sense of Difference
SDQ	Strengths and Difficulties Questionnaire
SE	Self Esteem
SEM	Structural Equation Modelling
SI +ve	Positive Self Image
SI –ve	Negative Self Image
SIP	Self Image Profiles
SIP-A	Self Image Profiles for Adolescents
SIP-C	Self Image Profiles for Children
SMR	Standardised Mortality Ratio
SRRS	Social Readjustment Rating Scale
STAI	State-Trait Anxiety Inventory
SUDEP	Sudden Unexpected Death in Epilepsy
WHO	World Health Organisation

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1.1 Stress

Stress derives from the Latin verb 'stringo' and formed part of the English language in the 14th century to convey an unpleasant condition of the environment.(Hayward 2005) The modern concept of stress is a response to a threatening situation with the aim being to maintain normal function. The response is however dependent upon the challenge caused to normal function, the person's perception of the situation and the perceived ability to cope with it. (Goldstein & Kopin 2007) The view that an organism will respond to a threat in order to maintain normal function stems from work undertaken by Cannon. He hypothesised that an organism placed under threatening conditions causes a physiological response in order maintain normal body function.(Le Moal 2007)

Selye advanced the field stress by suggesting that a wide variety of stimuli can be perceived as a threat that will then cause a stress response. He focused upon the biochemical response of stress and suggested that any demand, whether physical or psychological will cause the body to undergo the same process in order to readjust or adapt, the General Activation Syndrome' (GAS).(Patterson 1988) The GAS was described as initially causing a neuro-endocrine activation, similar to the physiological response observed by Cannon, followed then by tissue defence with the end result being exhaustion. (Selye 1998)

Selye's concept of prolonged stress being able to cause disease has been described as being widely accepted,(Goldstein & Kopin 2007) however it has been debated as to whether the body response to stress will be the same for all stimuli. This idea has been debated as it has been said that different physiological responses do occur in response to stimuli, with the response being dependent upon the emotion that it produced. Mason found that when fear and uncertainty were eliminated, GAS did not occur.(Le Moal 2007; Mason 1975)

The stress theory as presented by Selye focused upon a stimulus-response paradigm, however this was criticised, as it did not explain why different individuals experience stress in different ways. Theorists have attempted to explain the variance in the amount of stress experienced by either stating that it is due to the type of stimulus being

experienced, or by focusing upon identifying attenuating factors that affects the magnitude of stress.

Theorists such as Holmes and Rahe focused upon the cause of stress. They created a list of events and gave each a 'life change unit,' with a higher unit value given to those that they felt had the potential to impose more stress and subsequently more likely to cause stress.(Holmes & Rahe 1967) This method was advanced by Kanner et al to develop the 'Daily Hassles and Uplifts' scale to include everyday events,(Kanner et al. 1981) in order to appreciate that minor disruptions in everyday life had a potential to have an impact on health. A main disadvantage of focusing upon the cause of stress is that the list of potential stressful stimuli is endless. These models do imply that both large and small events have got the potential to cause stress. This is useful to understand, as a seemingly insignificant event may actually be the cause of significant stress to an individual. However by determining the likelihood and degree of stress that will occur, according to the type of event has been experienced, an assumption is made that the amount of stress experienced will be uniform. As mentioned by Horowitz, the subjective element of stress is lost. (Horowitz, Wilner & Alvarez 1979)

In order to account for stress being a subjective experience, other theorists have focused upon understanding the processes involved that determine the stress response. Theorists such as Lazarus and Folkman formed the Transactional Model of Stress and Coping(Lazarus & Folkman 1984) in order to highlight that there are more factors involved in a stress response than simply the stressful stimuli and the body's physiological reaction. They suggested that the occurrence of stress is dependent upon an individual's perception of the event. Lazarus and Folkman termed this appraisal; assessing the situation according to the issues that the event will create and their own ability to solve them. (Cox 1987; Dewe 1997; Leonova 1998) They emphasised that a cognitive appraisal of stressful stimuli is able to increase or decrease the amount of stress experienced by an individual, which determines how they are then able to adapt to the stressful event.(Lazarus 1993)

The Transactional Model of Stress and Coping introduces the role of mediating factors that can affect the outcome of stress. This is interesting as it is not contesting how much stress an event will cause, but rather that stress is dependent upon an individual feeling that they are able to cope with the demands and that they have the necessary resources to deal effectively with it.

So far, the different theories of stress that have been presented focus on stress being viewed on an individual level. This thesis is concerned on understanding stress when applied to a family, making it important to understand family stress theories. What was clear from the individual stress theories was that there was an initiating event that then had the potential to cause stress. This was especially salient in the stimulus theories as presented by Holmes and Rahe, with different events being posited as having causing varying degrees of stress. It will now be attempted to assess the different ways that researchers interested in the field of stress have attempted to identify what is specific about an event that makes it more likely to cause stress.

1.2 Stressors

The collective term for any factor that could generate stress is stressor. Stressors are conditions, experiences and activities that are viewed by the individual as a threat with the potential to become problematic.(Pearlin et al. 1990) Hill defined stressors as 'crisis-provoking events',(Hill 1958) with McCubbin and Patterson advancing the definition to include normative transitions, prior strains, intrafamily and social ambiguity.(McCubbin, Patterson & Sussman 1983) A stressor could therefore be any situation that can be viewed as the source of stress.

Kazak stated that the effect of a stressor varies between individuals due to events being perceived in different ways,(Kazak 1992) echoing the central premise of Lazarus and Folkman's stress theory that perception of an event determines if stress will occur. Thus different reactions arise with the same event making stressors a variable that is not uniform.(Hill 1958)

It has been attempted to identify the linking factor between all stressors. A principle thought has been whether the event causes an alteration to normal behaviour,(Hinkle 1974) with the potential to cause change being the underlying factor. (Boss 1992)

Upon reading the literature surrounding stress theories, there appears to be a conflict between stress theorists as to whether change is the factorial element that causes stress. Hill stated that a stressor can be identified when it produces a change within a system.(Hill 1958) This was based on his theory that crises within a family occur when a new situation presents itself, the underlying element being that it caused a change within the structure, roles or relationships of the family. Holmes and Rahe's list of stressful events were determined as they were all felt to cause change. (Holmes & Rahe

1967) However, Hobfoll et al criticised this as they argued that the events included could be interpreted as being both positive and negative situations. They felt only events that produced a negative change are stressful, as these situations pose a risk or potential loss.(Hobfoll & Spielberger 1992) Boss felt that change resulting in loss was most stressful when the loss was ambiguous.(Boss 1992) An example of an ambiguous loss could be living with a chronic condition or awaiting a diagnosis; when there is uncertainty surrounding the change in situation.

However, Kazak argues against change being the stressful element in stressors due to change being so inextricably related to stress and states instead that it is the degree of control an individual has over an event that determines whether stress occurs.(Kazak 1992)

From reviewing the differing opinions, it appears that a stressor is an event that demands a response. The response undertaken is then dependent upon how it has been interpreted by the individual. It has been suggested that likelihood of stress occurring from an event is also influenced by the individual's disposition at that particular time. Thus, if someone is already feeling stressed and an additional stressor presents itself, the amount of stress experienced will be greater as they have been sensitised.(Nixon & Bryant 2003) Patterson felt that an increase in stress experienced in this situation might occur due to fewer resources available to cope with future stressors. (Patterson 1988)

McCubbin and Patterson first introduced the concept of stressful events having a cumulative effect within their Double ABCX Model. They termed this situation as 'pile-up' of different stressor events, with each stressor presenting their own demand for change and increasing the strain upon the family.(Lavee, McCubbin & Patterson 1985)

The reaction to a stressor has been said to be different according to whether it is affecting an individual or an entire family. Hill felt that the likelihood of family stress was dependent on not only the stressor but also the family's ability to work together and use resources to deal effectively with it.(Hill 1958)

1.3 The Family

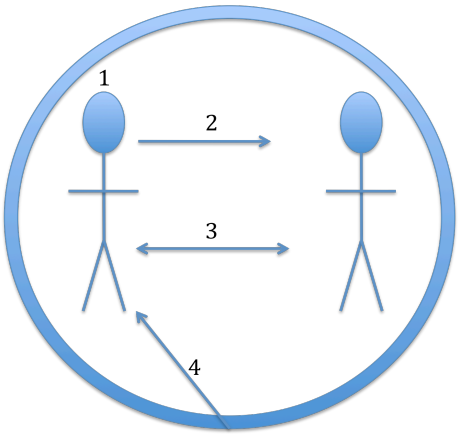
To understand stress in family, the meaning of family must be determined. A family consists of individuals who are united by a relationship based on a connection via blood, marriage, adoption or long-term commitment.(Patterson & Garwick 1994) Each individual within the family has a role that is assigned by their resigning community culture.(Hill 1958)

From an external view, the individuals within the construct of a family form a group, a unit that can be analysed as a whole rather than on an individual basis.(Kazak 1992)

When family refers to a single unit, ideas and interpretations do not represent those of an individual but of the family as a whole.(Patterson & Garwick 1994)

Lazarsfeld and Menzel suggested that within a family unit, each individual has four different types of properties.(Gilliss 1983) This is shown in Figure 1.

Figure 1: Properties of an Individual Within a Unit



Key

1 = Absolute: comes from the individual
2 = Relational: relation with other members
3 = Comparative: compare a member to another
4 = Contextual: Describes an individual according to the collective.

Figure 1 represents the properties within the family when viewed as an unit. The 'absolute' property represents the individual's characteristics, such as the symptoms of a disease they are suffering from. The 'relational' and 'comparative' properties represent the structure of the family; how the family functions and interacts with each other. The 'contextual' property is a global view of the unit, a static measure that appears to be similar to the theory of family meanings.(Gilliss 1983)

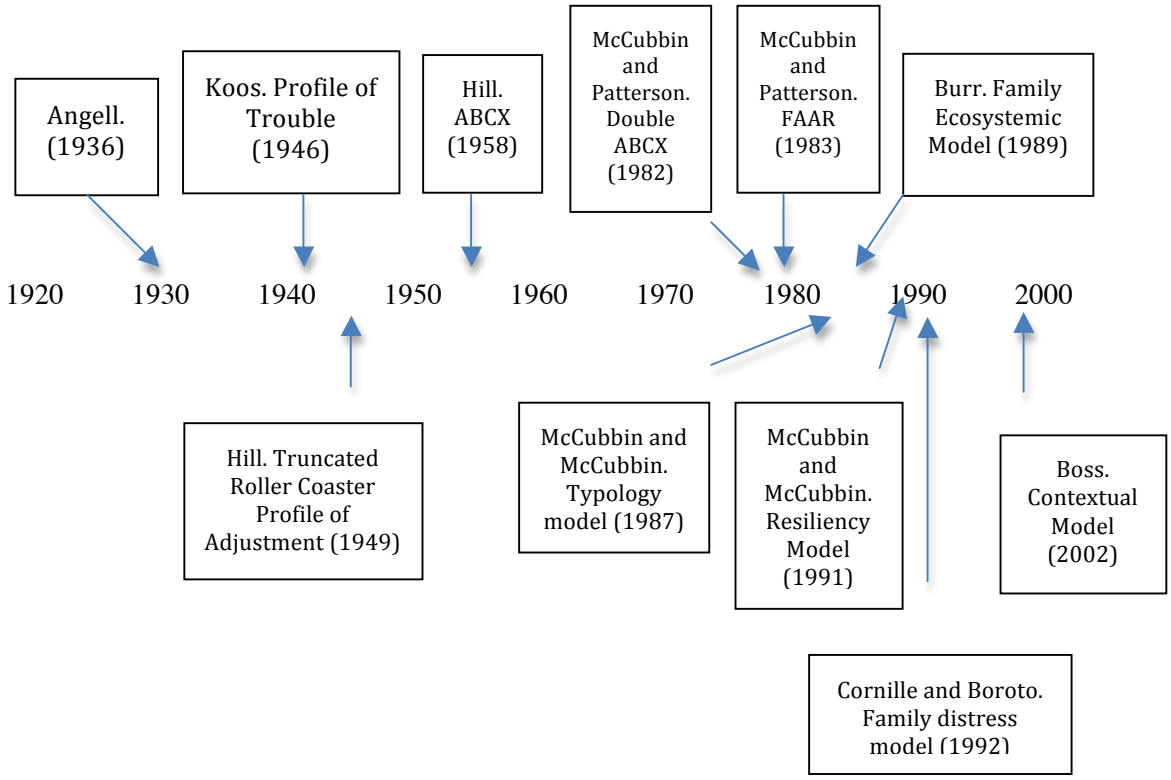
What is apparent from this diagram is that there are exchanges between family members, that family members are affected by and affect each other. It is these transmissions of behaviour and emotion between the family members, which suggest that the family has the ability to affect the health and wellbeing of its members.(Larson & Almeida 1999)

The emotional attachment between a parent and their child has been considered a fundamental component as in understanding how the parent feels stress when their child is diagnosed with a chronic condition. The parent and child relationship is additionally said to influence whether the child is able to adapt to the condition and cope effectively. (Wood, Klebba & Miller 2000) This makes it important to examine whether carers or parents of children do feel stressed when their child has been diagnosed with a chronic condition and if this has influenced the child's ability to adapt positively in response to the condition.

1.4 Family stress theories

Family stress was defined by McCubbin as the 'state which arises from an actual or perceived demand-capability imbalance in the family functioning'.(McCubbin 1983) Theories surrounding family stress have evolved over time. This can be displayed diagrammatically on the timeline below.

Figure 2: Timeline of Family Stress Theories, adapted from (Weber 2011a)



Boss named Reuben Hill as the father of family stress due to his creation of the ABCX model.(Boss & Mulligan 2003) However, interest into understanding how a family reacts to a sudden and dramatic change, stems from work carried out in the 1930s by Angell, examining the effect that the Great Depression had upon individuals and then the family as a unit.(Weber 2011a)

Koos developed the 'Profile of Trouble' and this was later expanded upon twice: Hill with the theory 'Truncated Roller Coaster Profile of Adjustment' and Burr 'Family Ecosystemic Model of Stress'.(Weber 2011b) These models present a linear view of the stress process, displaying the level of family function before, during and after the stressor event. Koos's and Hill's models display the different types of functioning after the stressor event, either by the level of interaction or adjustment. Burr's model

displays the different coping processes that a family can use so to adjust to the stressor. (Weber 2011b) These models represent a two-dimensional viewpoint of stress and do not attempt to explain the different variables involved in determining whether a family experiences stress. For this reason, the thesis will concentrate on the family stress models that are based upon Hill's ABCX Model, focusing on the ABCX Model, Double ABCX Model, Family Adjustment and Adaptation Response Model and the Resiliency Model of Family Stress, Adjustment and Adaptation.

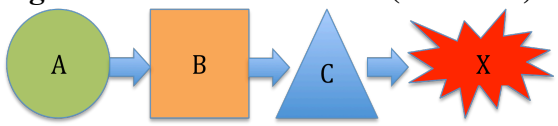
Family Stress Theories based on Hill

1.4.1 The ABCX Model

The majority of theories within family stress appear to be based on the Hill's ABCX Model formulated in 1958. Since then, the ABCX has remained at the core of many theories surrounding family stress, with models developed so to further help understand the variables involved before and after a stressful event.

The ABCX Model was proposed to explain the difficulties of family functioning when a family is subjected to stressors.(Hill 1958) The ABCX Model identifies a linear interaction of specific variables, with the outcome being that the family can be in a state of crisis. This is shown in Figure 3.

Figure 3: The ABCX Model (Hill 1958)



Key
A = Crisis precipitating event/stressor
B = Crisis-meeting resources
C = Family definition of the event
X = Crisis

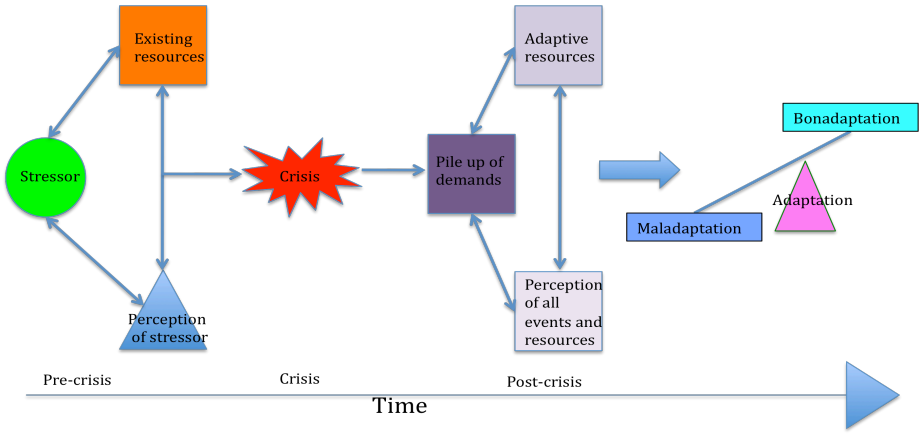
Within the model, 'A' denotes the stressor event, any situation that a family may view as being problematic.(Hill 1958) This may be a child's illness or an increase in condition severity. The 'B' signifies the family's existing resources that will allow them to adapt to the stressor. The resources may be the education and employment of the parents and the amount of support available. With plenty of resources, a family is more likely to form an organised response and are able to meet the demands created by the stressor.(Hill 1958) Hill also felt that a family's perception of the stressor event was important in understanding family stress, represented by 'C'. He felt that a family

was more likely to alter their normal behaviour if a stressor was viewed as being challenging.(Hill 1958) According to this model, 'A' interacting with 'B' interacting with 'C' produces a crisis, represented by 'X'.(Weber 2011c) In this context, a crisis refers to the negative consequences that occur when a family is presented with a stressor. (Hill 1958) An example for this would be the family becoming unsatisfied with their relationships and the altered family behaviour. A crisis is therefore a state of dysfunction.(Weber 2011c) As a crisis is an outcome of the stressor's effect on a family, it has been argued that a process of adaptation must also occur.(Lavee, McCubbin & Patterson 1985)

1.4.2 The Double ABCX Model

A primary criticism of the ABCX Model was that it did not include the events after the crisis had occurred, either to explain or predict how a family was to recover. To account for the post-crisis period, McCubbin and Patterson produced the Double ABCX Model in 1983. This can be seen in Figure 4. Their primary aim was to highlight that stress causes a multidimensional demand upon the family, by causing the family to adjust to the stressor event and to adapt their behaviour in order to meet the demands.(McCubbin 1983)

Figure 4: **The Double ABCX Model,** adapted from **(Lavee, McCubbin & Patterson 1985)**



The Double ABCX Model is divided into three sections: pre-crisis, crisis and post-crisis. The pre-crisis and crisis are essentially the same as Hill's ABCX Model. The

post-crisis period can be viewed by two entities; that each factor contributes directly to adjustment, and that each factor can attenuate the impact of the other factors considered in the post-crisis period altering the adjustment of the family to the stressor event.(Lavee, McCubbin & Patterson 1985; McCubbin 1983)

The post-crisis period introduces a pileup of stressors, to account for a single event causing additional new sources of stress. It considers that a family will use existing as well as newly attained resources and that a family will generate a new perception of all the events and available resources.(McCubbin 1983)

Within the Double ABCX Model, the outcome is adaptation, a description of the family's ability to balance the demands of the stressor(s) and their ability to meet the demands. This determines how family functioning has had to change. The change can either be negative (maladaptation) or positive (bonadaptation). (McCubbin 1983) Bonadaptation implies that the family has been able to balance the demands and capacity to cope with them. This is characterised by maintaining or strengthening family integrity and individual member's wellbeing. Maladptation implies that there is an imbalance between the demands placed on the family and their ability to meet them. When maladaptation occurs, family integrity and a family member's wellbeing deteriorate, which causes a negative effect on their physical and/or psychological health. (Lavee, McCubbin & Patterson 1985; McCubbin 1983)

1.4.3 The Family Adjustment and Adaptation Response (FAAR) Model

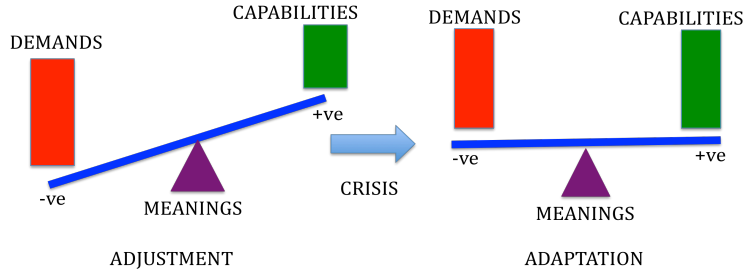
The Family Adjustment and Adaptation Response (FAAR) Model was developed by McCubbin and Patterson in 1983, with their Double ABCX Model incorporated into it. The FAAR uses the variables identified within the Double ABCX Model but places greater emphasise on the processes that a family undergoes in order to adjust and adapt to a stressor. They state that the two processes of adjustment and adaptation are central to understanding how a family responds to stress, both phases having independent aims to maintain a balance between demands and capacities so that a family is able to function normally. The adjustment phase is separated by the state of crisis before the process of adaptation can then occur.(Patterson 1989; Patterson & Garwick 1994)

A main element of the FAAR is that it views a family's response to a stressor over time, claiming that a family being presented with a stressor is able to go through cycles

of adjustment, crisis and adaptation rather than the linear description given by the Double ABCX Model. (Lavee, McCubbin & Patterson 1985; Patterson 1988)

After reading the literature on the FAAR Model, it appears that the adaptation process is largely the same as the post-crisis period of the Double ABCX Model, as the outcomes are again either positive (bonadaptation) or negative (maladaptation). These outcomes reflect whether the family has been able to restore a balance in their functioning by re-defining the stressor, reducing the pileup of demands or by developing new resources and coping strategies.(Patterson 1988) The difference of the FAAR Model compared to the previous models mentioned is that it emphasises that a process must occur before a family is in a state of crisis, termed adjustment. The outcome of adjustment are again either positive (bonadjustment) or negative (maladjustment), viewed according to whether family functioning and family member's health is positive or negative.(Patterson 1988; Patterson & Garwick 1994) Only if the family has been unable to meet the demands of the stressor, maladjusted, then a family will enter into a state of crisis. After the state of crisis, adaptation has the aim to restore family functioning by either reducing the demands and/or increasing the family's capabilities to deal with them. This can be seen diagrammatically in Figure 5. The 'demands' denote stressors, either being acute, chronic, major or minor stressful events. The 'capabilities' refer to the family's resources and coping behaviours that determines how the family will react.(Patterson 2002)

Figure 5: The Family Adjustment and Adaptation (FAAR) Model, adapted from (Patterson 1989)



As introduced by previous theorists, Patterson amended the FAAR Model to account for different families perceiving stressors differently. She introduced different levels of meanings according to the family's situation, the family's unit identity and the community that the family resides in. These different meanings represent the underlying reason as to why the stressor has the potential to cause stress to the family.(Patterson & Garwick 1994)

Using the example of a chronic illness being diagnosed within a family, the situational meaning may cause the family to be in denial with the diagnosis. This is due to the chronic illness diagnosis not relating to the family's definition of children being healthy. The family level meaning of the chronic illness diagnosis may make their normal routines become disrupted, as the family has to restructure itself to meet the demands of the chronic condition. The global view meaning can cause the family to feel uncertainty about the future or feel that they are stigmatised by the community.(Patterson 1988; Patterson 1989; Patterson & Garwick 1994)

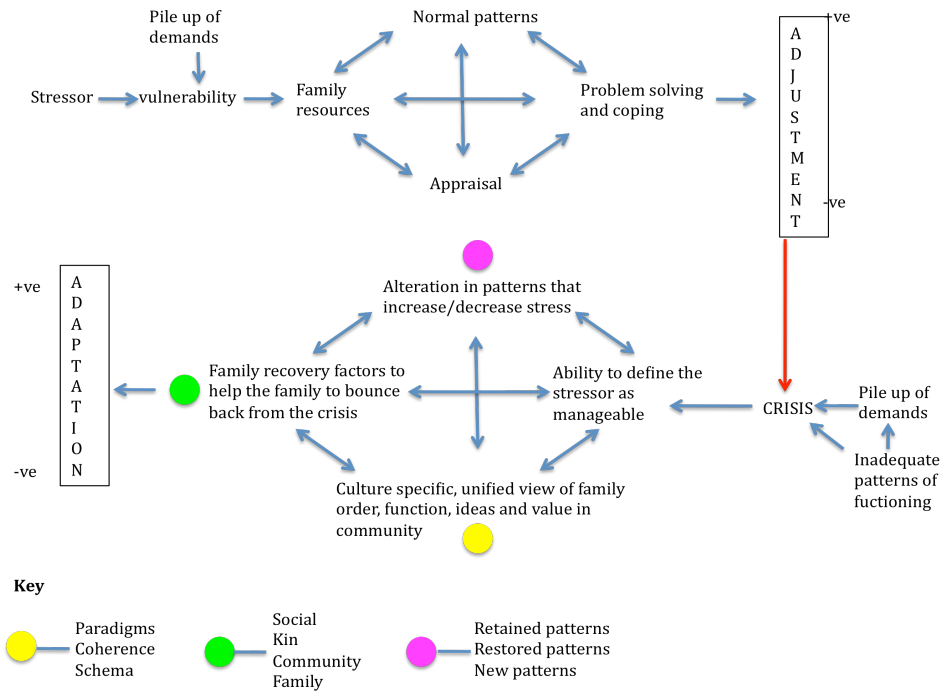
Together, these levels of meanings determine the behaviour and emotion expressed by the family when presented with a stressor event, in turn influencing whether the family is able to bonadapt.

1.4.4 The Resiliency Model of Family Stress, Adjustment and Adaptation

The Resiliency Model of Family Stress, Adjustment and Adaptation was developed by McCubbin and McCubbin and underwent various modifications from 1993 to 2001. (Weber 2011d) It was created to identify specific methods that a family may employ when attempting to maintain family functioning whilst adjusting and adapting to a stressor. It uses the same progression of events as identified within the FAAR Model, that there is a period of adjustment which can then progress to a crisis event requiring a period of adaptation to restore family functioning. The emphasis of the Resiliency model is within the post-crisis adaptation phase, expanding upon the variables within the Double ABCX Model so to explain why some families are resilient and are able to recover from a crisis. (McCubbin et al. 1997) (Smith 1999)

The Resiliency Model does not add to the understanding of the family stress process, using the processes described within the FAAR Model but gives a more detailed explanation of the specific family capabilities that aid a family's recovery from a stressor. This can be seen in Figure 6.

Figure 6: The **Resiliency Model of Family Stress, combining the Adjustment and Adaptation Phases.** Adapted from (Weber 2011d)



The areas that the Resiliency Model state are important in determining whether a family is able to adapt positively to a stressor and subsequent crisis are: the patterns in family functioning (retained, restored or newly initiated patterns), the resources of support used by the family (family, social, kin and community support), ability to problem solve and cope as well as the meaning that the family gives to the event (how family should function, is the family able to manage). (McCubbin et al. 1997) The different factors involved in the model conceptualise that resiliency is multifactorial and that successful methods employed will differ according to the situation, the aim being to restore balanced family functioning. (Masten, Best & Garmezy 1990; Patterson 2002)

1.5 Summary of the Family Stress Theories

The different theories involved in family stress have been in attempt to explain how families react to stressful events. A comparison of the sequence of events described within the four family stress models presented within this chapter can be seen in Table 1.

Table 1: Comparison of the Examined Family Stress Models. Adapted from (Weber 2011e)

ABCX	Double ABCX	FAAR	Resiliency
Crisis-precipitating event	Pre-crisis: stressor	Adjustment phase: stressor	Adjustment phase: stressor
		Adjustment phase: demands	Adjustment phase: vulnerability, pileup of demands, normal patterns of functioning
Family's crisis-meeting resources	Pre-crisis: existing resources	Adjustment phase: existing resources	Adjustment phase: family resources
Family's definition of the event	Pre-crisis: perception of stressor	Adjustment phase: definition and appraisal of demands, coping strategies	Adjustment phase: stressor appraisal, problem solving and coping
		Bonadjustment or maladjustment	Bonadjustment or maladjustment
Crisis	Crisis	Crisis-maladjustment	Crisis-maladjustment
	Post-crisis: Pileup	Adaptation phase: pileup	Adaptation phase: pileup, inadequate +-deteriorating family functioning
			Adaptation phase: patterns of functioning-retained, resorted or new
	Post-crisis: existing and new resources	Adaptation phase: resources and support	Adaptation phase: family resources-social, family, community and kin support
	Post-crisis: perception of all events	Adaptation phase: situational appraisal, agree on solutions, shared meaning	Adaptation phase: situational appraisal-paradigms, coherence, schema
	Post-crisis: coping	Adaptation phase: restructuring and adaptive coping	Adaptation phase: Problem solving and coping
	Bonadaptation or Maladaptation	Bonadaptation or Maladaptation	Bonadaptation or Maladaptation

As seen within Table 1, the theories have become more complex as they have evolved over time in order to appreciate the many factors that are involved in the process of family stress and to emphasise that it is a non-uniform response. It appears however that the underlying assumption of the different models in family stress is that an imbalance between the demands and a family's capability to meet them ultimately

results in family stress. With each family stress model, there are strengths and weaknesses. These can be seen in Table 2.

Table 2: Strengths and Weaknesses of the Examined Family Stress Models

	Salient points of the model	Strengths of the model	Weaknesses of the model
ABCX	Creates a formula to conceptualise the process of stress with crisis being the outcome.	Identifies key factors: stressor, resources and perception.	Linear progression with only outcome being crisis. No explanation of what happens after a crisis.
Double ABCX	Has a pre- and post-crisis period. Post-crisis period explains the structural components and sequence of adaptation that are involved in the variability of adaptation.	Identifies a post-crisis period and that a family is able to adapt to a stressor when it has caused a crisis. Highlights coping.	Implies that a crisis will occur when presented with a stressor. Only describes the relationship between the components rather than the processes to explain and predict how families respond to stress.
FAAR	Portrays family adjustment and adaptation to a stressor over time. Emphasises that not all families will progress to a state of crisis due to their ability to adjust with minimal change needed. States that the underlying process is a balance between demands of stressor and capabilities of a family to meet them.	Highlights the dynamic process of stress: the processes involved when presented with a stressor and the reason why there is variability in different families responses. Integrates coping as a resource before a crisis occurs rather than simply a post-crisis event.	It is not specified as to what the resources are that assist in a family's capability to meet the demands made by a stressor.
Resiliency	Expands upon the post-crisis adaptation phase by identifying key factors that help to explain why some families are resilient to crisis whilst others are not able to adapt.	Specifies the specific demands and capabilities. Makes it clear that family functioning, shared meaning, support and ability to problem solve and cope, are important in making a family resilient.	Detailed model with different types of situational appraisal that may be useful theoretically but difficult to ascertain in a practical setting.

The ABCX Model introduced the concept that when a family was subjected to a stressful event, the available resources to meet the demands and the family's definition of the event were important in determining if it is a threat to their family function. The Double ABCX Model applied this premise but indicated that additional factors needed

to be considered, as they were involved in a family's recovery from the event. Both the ABCX Model and the Double ABCX Model suggested that a progression of events that lead to a crisis. The FAAR Model altered the view that all stressors will result in a crisis by including the adjustment phase whereby minor stressful events can be managed without causing much change to the family. The Resiliency Model helped to specify the main factors that assist in a family's capability to meet the demands that originated from the initial stressor event.

Reviewing the theories surrounding family stress helps to describe and ultimately predict the link between the family system and the health and illness in an individual family member.(Patterson 1988) Using the Double ABCX, FAAR and Resiliency Model, maladaptating to the initial stressor event causes imbalance in the family system, at the detriment of family structure, function and family member's wellbeing. The theories appear to have arisen due to research showing that a stressor does not categorically cause stress. Therefore, it cannot always be assumed that a child presenting with a potential diagnosis of a chronic condition will cause the parent to be stressed. The theories do however state that a pile-up of stressors may occur, making it salient to examine for minor stressors such as subjective parental perception of their child's characteristics, for example behaviour. The theories additionally highlight that the capabilities of a family to respond to a stressor are vital in their ability to adapt. The Resiliency Model in particular states that family functioning, coping and support resources are all factors that aid adaptation. This makes it important to examine these factors in future research on family stress.

2.1 History of Epilepsy

Epilepsy is regarded as one of the oldest conditions known to mankind.(de Boer 2010) Rajendra Kale described the history of epilepsy as '4000 years of ignorance, superstition and stigma, followed by 100 years of knowledge, superstition and stigma'.(Kale 1997) This statement highlights the misconceptions of epilepsy that have been present throughout history and that are still thought of in the 21st century.

Humans suffering with seizures have been documented in different civilisations, with the first recorded epileptic seizure thought to be in 2000 B.C. in Mesopotamia.(Magiorkinis, Sidiropoulou & Diamantis 2010) Ancient Egyptians, Romans and Greeks all documented epileptic seizures, lending the Greeks to coin 'epilepsy', taken from the verb meaning to seize. (Magiorkinis, Sidiropoulou & Diamantis 2010)

The attitude towards epilepsy alters according to the cultural beliefs, the geographical location and the particular era. At certain times in history, to have the tendency for epileptic seizures was an attribute held in high esteem, a belief held in Senegal. However, the prevailing concept of epilepsy throughout history is negative, often linked to demonic forces, witchcraft and psychological disease.(de Boer 2010)

The ancient Greeks connected epilepsy with superstition, viewing a seizure as a punishment to sinners. It was felt that the seizure type represented the god that was angry with them.(Magiorkinis, Sidiropoulou & Diamantis 2010) The connection between epilepsy and religion was also present throughout the Middle Ages, with seizures being viewed as a form of punishment.(Magiorkinis, Sidiropoulou & Diamantis 2010),(Riggs & Riggs 2005)

It was Hippocrates of Kos in his text 'On sacred diseases' that first disputed the divine origin of epilepsy, and instead classed it scientifically. He stated that the aetiology of epilepsy was due to a brain dysfunction rather than a condition that arose from magical or divine causes.(Magiorkinis, Sidiropoulou & Diamantis 2010)

The father of modern epilepsy is considered to be John Jackson due to his modern approach of combining clinical history and neurological examination to determine the localization of the epilepsy.(Loring 2010) He further described epilepsy in 1873 as an

'occasional, sudden, excessive, rapid and local discharges of gray matter', a physiological definition that is still used today.(Akimoto 2004) However, even during the early 20th century, those with epilepsy were often segregated from society and were placed in asylums, due to the belief that it was a contagious disease and linked with lunacy.(Sander, Barclay & Shorvon 1993)

2.2 Definition of Epilepsy

Jackson accurately identified and described the physiology of epilepsy in 1873, a description of the phenomenon of abnormal and excessive excitability of cortical neurones within the cerebral hemispheres. (Akimoto 2004; Appleton & Gibbs 2004d) It is the synchronous paroxysmal neuronal discharges that originate from the cortex that produces the clinical manifestation of recurrent and unprovoked seizures (photosensitivity is the only exception). (Fisher et al. 2005), (Banerjee, Filippi & Allen Hauser 2009)
The International League Against Epilepsy (ILAE) defines epilepsy as

'A disorder of the brain characterised by an enduring predisposition to generate epileptic seizures and by the neurobiologic, cognitive, psychological and social consequences of this condition.' (Fisher et al. 2005)

The ILAE further states that for epilepsy to be diagnosed, the occurrence of at least one epileptic seizure must occur.(Fisher et al. 2005) It is more usual for epilepsy to be diagnosed after the person has had two unprovoked seizures as this strengthens the claim that the individual has a tendency for epileptic seizures.(Arts & Geerts 2009; Banerjee, Filippi & Allen Hauser 2009) An epileptic seizure is the end product of the cortical discharges, producing symptoms that can be either sensory, motor or autonomic clinical manifestations.(Fisher et al. 2005) The ILAE defines an epileptic seizure as 'a transient occurrence of signs and/or symptoms due to abnormal excessive or synchronous neuronal activity within the brain.' (Fisher et al. 2005) An epileptic seizure causes stereotypical disturbance of one or several brain functions such as consciousness, behaviour, emotion or sensation and it is this disturbance that often

makes epilepsy clinically obvious, however, the alteration can be subtle and not easy to recognise. (Appleton & Gibbs 2004d)

2.3 Diagnosis

Recognising the epileptic seizure is essential for epilepsy to be diagnosed. This is because a diagnosis of epilepsy is achieved by clinical evaluation, guided by a detailed history of events leading before, during and after the suspected epileptic seizure.(Elger & Schmidt 2008) The failure of obtaining an adequate history can lead to misinterpretation of clinical signs and this increases the risk of falsely diagnosing the individual as having epilepsy.(Gibbs & Appleton 1992) It has been stated that the diagnosis of epilepsy should be considered at four separate levels: (Appleton & Gibbs 2004d)

1. Recognition of epileptic seizures
2. Classification of seizure type
3. Identification of epilepsy syndrome
4. Determination of aetiology

A neurological examination should always occur as an abnormal examination is said to predict seizure recurrence. (Stokes et al. 2004) However, clinical examinations are often normal, making an accurate history taking of the event even more vital. (Appleton & Gibbs 2004d)

2.4 Epidemiology

The epidemiology of epilepsy has been reviewed through descriptive and analytical studies. The World Health Organisation (WHO) described epilepsy as a condition that imposes physical, psychological, social and economic burdens on individuals, families and countries.(WHO 2005) The WHO recently stated that epilepsy contributed 1% towards the global burden of disease,(WHO 2005) a burden-specific measure based on epidemiological rates and years of life lost due to ill-health. This figure helps to represent the health problem created by epilepsy for not only the individual with epilepsy, but also their families and society.(de Boer 2010)

Epilepsy is common worldwide with the WHO predicting that around 50 million people worldwide have epilepsy (WHO 2005) with 3% of any population having had epilepsy at some point within their lifetime.(Berkovic et al. 2006) The prevalence rate of active epilepsy is quoted within the range of 5-10 per 1000 people, (Sander 2003) with an incidence rate in developed countries estimated to be between 40-70 per 100'000 of the population per year.(Sander 2003) The distribution of epilepsy is non-uniform, with the incidence of epilepsy being higher in young children and older people.(Carpio & Hauser 2009) (Kotsopoulos et al. 2002)

Epilepsy is able to resolve itself, with remission more likely to occur in new onset cases of epilepsy as the shorter the length of time experiencing seizures, the more likely that they will respond to treatment. (Shorvon & Luciano 2007) The likelihood of remission from seizures has also been found to be dependent upon the number of seizures experienced within the first 6 months of initial presentation; fewer seizures experienced decreases the length of time taken to reach a remission.(Lhatoo et al. 2001) (Shorvon 1984; Sillanpää & Schmidt 2006)

The impact of being diagnosed with epilepsy has the potential to cause psychosocial consequences due to inability to drive and causing a disruption to education.(Wiebe et al. 2009) Epilepsy is not only linked to causing psychosocial consequences, but is also related to increased mortality. An epileptic seizure can itself cause death, with this being a primary fear for parents especially when they witness their child's first seizure.(Wiebe et al. 2009) The standardised mortality rate increases by 2-3 times in patients with epilepsy when compared to the general population.(Shorvon & Luciano 2007) The United Kingdom National General Practice Study of Epilepsy (NGPSE) revealed a standardised mortality ratio (SMR) of 2.1; for every expected death there was an additional death.(Lhatoo et al. 2001) When epilepsy is newly diagnosed, the rise in mortality is due to its underlying cause, whilst in chronic epilepsy the excess is often due to the event of the seizure: accident or sudden unexpected death in epilepsy (SUDEP). (Lhatoo et al. 2001; Shorvon & Luciano 2007) The NGPSE also stated that a higher mortality was seen in younger patients with chronic epilepsy, reasons attributed to SUDEP and other epilepsy-related causes of deaths.(Lhatoo et al. 2001)

2.5 Aetiology

The aetiology of epilepsy varies widely in relation to geographical location and different risk factors are associated with different age groups. It is also thought that the aetiology of epilepsy is multifactorial; interaction between infectious agents, social, geographic, genetic, toxic and environmental factors all contributing to the risk of developing epilepsy.(Sander 2003) This makes the potential list of causative factors extensive.

Despite this, for around 70-75% of individuals who are diagnosed with epilepsy, no specific cause may be found.(Appleton & Gibbs 2004a; Berkovic et al. 2006) It has been further hypothesised that an estimated 40% of those who have epilepsy occurs due to a genetic predisposition.(Turnbull et al. 2005)

The different causes of epilepsy have been suggested by the ILAE to be grouped under the following terms:(Berg et al. 2010)

1. Structural/metabolic
2. Genetic
3. Unknown cause

2.6 Investigations

As stated previously, epilepsy is a clinical diagnosis, therefore the aims of undertaking further investigations are to classify the epilepsy type or syndrome, identify the aetiology and for guidance in providing the most effective management plan.(Appleton & Gibbs 2004e) The investigations that can be ordered to assist the clinician in these areas are: (Stokes et al. 2004)

- Electroencephalography (EEG)
    - Standard EEG- awake with photic stimulation and hyperventilation over 30 minute duration to increase cortical excitability.
    - Sleep EEG- sleep deprivation is known to increase cortical excitability, increasing the likelihood of an epileptic seizure to be identified on an EEG. (Badawy et al. 2006; Civardi & Collini 2007)
    - Ambulatory- EEG recording over a long period of time, increasing the likelihood of the seizure 'captured'

- Neuroimaging
    - Magnetic Resonance Imaging (MRI) is the neuroimaging of choice, as stated by the latest National Institute of Clinical Excellence (NICE) guidelines and the ILAE (Berg et al. 2010; Stokes et al. 2004)
- 'Other'
    - An extensive range of haematological, metabolic and genetic investigations can be performed. These should be considered for myoclonic epilepsy and neurodegenerative disorders that have epilepsy as a feature. (Appleton & Gibbs 2004e)

An EEG is an investigation that involves electrodes being placed onto the scalp surface with the aim being that it can record the epileptic neuronal discharges. Epilepsy is the collective name for a heterogeneous group of conditions that produce unprovoked, paroxysmal neuronal discharges.(Cowan 2002) The pattern of the discharge differs with different types of epilepsy, thus making the EEG a useful tool for classification. (Appleton & Gibbs 2004e) It should be noted however that an EEG is said to be normal for half of those with epilepsy as the sensitivity rate of an EEG is 50%. (Binnie & Stefan 1999) An EEG can also indicate the underlying cause of epilepsy by identifying structural brain lesions,(Binnie & Stefan 1999) however neuroimaging is the investigation of choice for identifying structural abnormalities.(Gaillard et al. 2009) When neuroimaging is ordered, NICE states that MRI should be the imaging investigation of choice.

Guidance on when to order an MRI is if: (Stokes et al. 2004)

1. Epilepsy has developed before 2 years of age or during adulthood
2. Focal seizure onset
3. Seizures continue despite first-line medication.

2.7 Classification

Epilepsies vary according to the causative factors and the pathophysiological mechanisms that underlie different types of epileptic seizures.(Badawy et al. 2007) The location of the neuronal discharge allows a seizure to be broadly classified as being generalised or focal type seizure. Focal seizures represent those with neuronal discharges that involve one cortical hemisphere at onset, however it is possible to then

spread and involve both hemispheres.(Badawy et al. 2007; Berg et al. 2010) Neuronal discharges in generalised seizures rapidly spread across engaging networks to involve both cortical hemispheres.(Berg et al. 2010; Berg & Cross 2010)

According to the seizure's physical manifestation, the seizure can be further classified according to the 2010 ILAE classification system.(Berg et al. 2010; Berg & Cross 2010) This can be seen in Table 3. As seen in Table 3, seizures can present in different ways and can make diagnosis an uncertainty, with studies reporting misdiagnosis rates of 4.6-30% in different settings. Often, this is due to alternative diagnoses being suggested, increasing the risk of inappropriate treatment. (Chowdhury, Nashef & Elwes 2008)

As well as being able to classify the seizure, it is sometimes possible to classify an epilepsy syndrome according to the clinical manifestation, EEG characteristics and the age of onset.(Berg et al. 2010) However between 30-40% of childhood epilepsies a syndrome may not be identifiable.(Berg et al. 2000) The importance of diagnosing an epileptic syndrome is because of the implications it has for predicting the individual's prognosis and likely aetiology as well as determining the most suitable management for them. (Appleton & Gibbs 2004b)

Table 3: Classification of epileptic seizures(Berg et al. 2010)

Classification of seizures			
Generalised	Tonic-clonic		
	Absence	Typical	
		Atypical	
		Absence with features	Myoclonic absence
			Eyelid myoclonia
	Myoclonic	Myoclonic	
		Myoclonic atonic	
		Myoclonic tonic	
	Clonic		
	Tonic		
	Atonic		
Focal	Without impairment of consciousness	Motor or autonomic components	
		Sensory or psychic phenomena	
	With impairment of consciousness	Dyscognitive	
		Evolves to a bilateral, convulsive seizure.	
Unknown	Epileptic spasms		

2.8 Management

The primary aim of managing epilepsy is to control seizures so that the individual can maintain a good quality of life.(Elger & Schmidt 2008) The mainstay treatment option

is starting an anti-epileptic drug (AED) that is taken on a daily basis as a preventative measure.(Stokes et al. 2004) Arts et al stated in a recent review that it is generally considered not necessary to start an AED treatment regime after a single unprovoked epileptic seizure. The reason being that there is a 30-70% chance of a child not having another seizure, thus making the risks associated from AED side effects outweigh the benefits.(Arts & Geerts 2009) However if an individual has had recurrent seizures, NICE recommends after two epileptic seizures, or equally if they are having a negative impact on the individual's quality of life, starting an AED would generally be recommended. (Appleton & Gibbs 2004c; Elger & Schmidt 2008; Stokes et al. 2004)

The aim of starting an AED is to 'achieve complete seizure control without unacceptable side effects'. (Appleton & Gibbs 2004c) For approximately 65% of patients, AEDs provide satisfactory seizure control in new-onset epilepsy.(Elger & Schmidt 2008) However, around a third of all patients do not respond to AEDs.(Elger & Schmidt 2008; Sisodiya 2007) If the first line agent fails to achieve control or the side effects were unacceptable, then a different drug should substitute the previous medication or to use the new AED as an add-on, with the recommended aim being monotherapy.(Appleton & Gibbs 2004c; Stokes et al. 2004)

There is a wide range of different AEDs available, with first line medications based upon clinical grounds and a careful individual risk-benefit assessment.(Elger & Schmidt 2008) The most recent NICE guidelines (2004) state that for children, sodium valporate should be started initially for generalised epilepsy and carbamazepine for focal seizures. A large randomised controlled multi-centre trial, the Standard and New Antiepileptic Drugs (SANAD) study, agreed with NICE that sodium valporate was the most suitable first line AED for generalised seizures,(Marson et al. 2007b) but suggested that lamotrigine should be the first line AED for focal seizures, due to its tolerability being better than carbamazepine.(Marson et al. 2007a) Regardless of the chosen AED, all are associated with side effects, with all holding the potential to cause a detrimental effect on cognition or behaviour.(Raspall-Chaure, Neville & Scott 2008) The risk of developing long-term consequences increases with chronic AED therapy and polypharmacy, especially relevant in children as this can restrict their academic achievement.(Loring & Meador 2004) This makes AED toxicity being an important preventable aspect of disability and poor health associated with epilepsy.(Raspall-Chaure, Neville & Scott 2008)

2.9 A Literature Review of the Studies Examining Stress in Parents or Carers of Children with Diagnosed Epilepsy

"People with epilepsy do not live in a vacuum: any negative consequences experienced are likely to extend to all family members"-Thornton and Upton (Ellis, Upton & Thompson 2000)

2.9.1 Carer Stress when their Child is Diagnosed with Epilepsy

Epilepsy is a chronic condition, described as such due to having a long duration with the potential to remain present over a life span, or that it may take time to resolve. (Duffy 2011; Mattsson 1972)

Chronic conditions are frequently associated with periods of acute exacerbations that require a greater intensity of medical attention.(Mattsson 1972) Epilepsy presents clinically with seizures, each one representing a potentially stressful event. As seizures have a likelihood of recurring, the number of stressful events that an individual is subjected to increases. (Mattsson 1972) It is the occurrence of seizures after a relative period of stability that makes the natural history of epilepsy to be described as being relapsing-remitting.(Sillanpää & Schmidt 2006)

A diagnosis of a chronic condition is known to be stressful for that individual along with their family as a whole, as it causes emotional distress throughout all members.(Holmes & Deb 2003; LoBiondo-Wood, Williams & McGhee 2004) Epilepsy has been shown to identify this relationship, with previous research has displayed that a diagnosis of epilepsy causes a negative impact on the health of those who are diagnosed with the condition as well as their family members. (Buelow et al. 2006; Hoare 1993; Hoare & Kerley 1991)

A normal role of being a parent to a child is being a caregiver.(Pearlin et al. 1990; Raina et al. 2004) However, if their child is diagnosed with a chronic condition such as epilepsy, it is more likely that the child's needs may exceed the usual needs of children and thus a higher level of care will be required.(Raina et al. 2004) As more care is required, dependency on the caregiver may increase. This may result in a restructured

relationship between the carer and recipient, as caring for the other person supersedes all other components of the relationship.(Pearlin et al. 1990)
The relationship between caregiving and health is often defined by the amount of stress that they are feeling.(Raina et al. 2004) Mash and Johnston defined parenting stress 'as a complex construct involving behavioural, cognitive and affective components'.(Solem, Christophersen & Martinussen 2011) Abidin describes parenting stress as an 'appraisal of the benefits and harm' that the parent undertakes with each situation that occurs to them whilst within the parenting role.(Abidin 1992)

Stress in parents of children with chronic conditions is a widely reported phenomenon.(Buelow et al. 2006; Dewey & Crawford 2007; Howe et al. 1993; Plant & Sanders 2007; Wallander & Varni 1998) This makes it unlikely for epilepsy to not also identify this relationship. A large pooled data set from the Centres for Disease Control and Prevention involving 78305 children and their parents, revealed that 44% of parents whose child had a developmental problem, with epilepsy being one such problem, experienced stress compared to 11% of stress being reported in parents whose child does not have any health needs. [120] Chiou et al compared stress in parents who have a child diagnosed with epilepsy against a control group involving parents whose child had been diagnosed with asthma, finding that parental stress was reported more frequently in parents who have been diagnosed with epilepsy.(Chiou & Hsieh 2008a)

Although it may be considered that all children diagnosed with a chronic condition may cause increased parental stress, epilepsy is a condition that has been described as producing unique challenges to parents who care for a child diagnosed with it.(Duffy 2011)
An important element of epilepsy-related parental stress is that seizures are unpredictable.(Duffy 2011; Modi 2009) Studies of parents who have a child with intractable epilepsy, reported that 45-65% of parents experienced stress when caring for their child.(Cushner-Weinstein et al. 2008; Murray 1993; Wirrell et al. 2008) Murray identified 41 parents who have a child with intractable epilepsy. The study findings revealed that many parents felt uncertainty as a result of not knowing whether a seizure was occurring or when the next seizure would occur.(Murray 1993) Nolan et al supported this finding in a study of parents of children with Dravet Syndrome, with

uncertainty being the main cause of stress to parents.(Nolan, Camfield & Camfield 2008) The uncertainty was told to recur episodically and this was suggested to relate to the unpredictable nature of epilepsy.(Cushner-Weinstein et al. 2008; Murray 1993) It should be noted that intractable epilepsy is a form that causes individuals to have many seizures making seizure reoccurrence a regular phenomenon. This means that it is likely that the reported stress levels in parents of children with intractable epilepsy will be higher than parents whose child has an occasional seizure.(Modi 2009)
Mitchell et al found during a longitudinal study of 119 children with epilepsy that seizure severity was significantly associated with more stress being experienced by the parent.(Mitchell, Scheier & Baker 2000)

Murray stated that parents of children with epilepsy expressed concern regarding their child's future with it being a pressing issue, and further found it to be related to increased anxiety in parents.(Murray 1993) In a literature review by Austin, parents and children were reported to be fearful of other people's responses to epilepsy due to their apprehension of being rejected and negative attitudes that would be expressed.(Austin 1996) Another literature review also found that stigma, regardless of whether it was actually encountered, was the biggest challenge that those diagnosed with epilepsy felt they had to face.(Austin et al. 2004b) However, a study into the prevalence of felt stigmatisation in adolescents by Westbrook et al, revealed that 66% of the 62 adolescents had never felt stigmatised with 33% reporting to have perceived some form of stigmatisation.(Westbrook, Bauman & Shinnar 1992)
Another reported seizure-related fear was mothers fearing death of the child.(Buelow et al. 2006; Hoare & Kerley 1991) Hoare et al reported that 31% of mothers stated fear of death out of a sample of 35 mothers of children who were having seizures.(Hoare & Kerley 1991) Cushner-Weinstein reported that parents felt incompetence in their ability to manage their child's seizures, decreasing their confidence when performing the parental role with the result being increased parent stress. (Cushner-Weinstein et al. 2008)

In order to portray factors that are associated with increased stress in parents who have a child with epilepsy, Buelow et al conducted semi-structured interviews with 20 parents who had a child diagnosed with epilepsy and an intellectual disability. She

identified factors that were reported as causes of stress but also highlighted the situation of 'pile up' stressors as identified within the Double ABCX Model.(Buelow et al. 2006)

The paper identified five broad sources of stress:

1. Concern with the child (future, behaviour and seizure consequence concerns)
2. Communication between family and healthcare providers (medication concerns, information need and length for epilepsy to be diagnosed)
3. Alteration in family dynamics (parent, sibling relationships, leisure activities)
4. Interactions with the child's school (communication, transition from school and safety issues)
5. The amount of support provided by the community for the family (family counselling, time off work and respite care)

These factors identified via semi-structured interviews have been found in other studies examining stress in parents who have a child with epilepsy.

The factors identified by Beulow display that a diagnosis of epilepsy has the potential to affect parental ability to maintain family function, with parental stress considered a mediating factor between family functioning and parenting variables.(Abidin 1992; Duffy 2011; Modi 2009; Rodenburg et al. 2007) The reason why family functioning should be considered when examining parental stress is first due to understanding the factors that may be contributing to stress and also due to family functioning being considered a part of the child's adjustment to epilepsy.(Austin 1996; Austin et al. 2004b; Rodenburg et al. 2007)

Examining the implications of parenting stress and the factors that contribute towards it helps to link family stress theories to a clinical setting. Different studies examining the impact of epilepsy on the family have used a family stress theory as the basis of their methodology. Austin(Austin 1996) and Rodenburg(Rodenburg et al. 2007) both used the Double ABCX model as the basis of their studies methodological rational and Mu(Mu 2005) has used the resiliency model for the same purpose. Studies assessing the impact that other conditions have on the family have also used the Double ABCX

model(Saloviita, Italinna & Leinonen 2003) and the resiliency model(Doucette & Pinelli 2004).

Identifying that other studies have used family stress theories as the basis of their methodology helps to strengthen the rational that parental stress has the potential to affect a variety of family life aspects. This is informative as it highlights the different variables that should be assessed within this present study.

The next part of this literature review will examine the factors that contribute to parental stress.

2.9.2 The Effect on a Carer when their Child is Diagnosed with Epilepsy

Carer Health

A child being diagnosed with a chronic health condition has been reported to cause mental health problems in parents and this has also been found in parents of children with epilepsy.(Ellis, Upton & Thompson 2000) (Dumas, Gibson & Albin 1989; Holmes & Deb 2003; Rodenburg et al. 2005; Shore et al. 2002) Depressive symptoms in parents, particularly mothers, have been reported when they care for a child diagnosed with epilepsy.(Austin et al. 2004a; Ferro et al. 2011a; Ferro et al. 2011b; Mu, Kuo & Chang 2005; Rodenburg et al. 2007; Rodenburg et al. 2005; Shore et al. 2002) Shore et al stated that depression may be more likely to occur in mothers caring for a child with epilepsy due to the demands placed upon them whilst caring for a child with a chronic condition, the stigma associated with seizures and the high rate of behaviour problems.(Shore et al. 2002) Other studies have found that parental depression resulted from uncertainty and ambiguity that surrounded the diagnosis of epilepsy.(Mu 2005; Mu, Kuo & Chang 2005; Mu 2008b)

Thus it could be concluded that it is the demands and subsequently the stress caused by these factors that contribute to the likelihood of depression occurring in parents. This was found in a study conducted by Rodenburg et al with their findings showing an association between parental depression and parental stress.(Rodenburg et al. 2007)

Shore et al found from 115 mothers that depression in mothers of children with epilepsy was significantly associated with low income, low satisfaction with family relationships and higher levels of behaviour problems.(Shore et al. 2002) The association between parental distress along with decreased mental health and child

behaviour problems appears to be a widely reported finding when the child is diagnosed with epilepsy.(Austin & Caplan 2007; Austin et al. 2004a; Ferro et al. 2011a; Wirrell et al. 2008)

The importance of studying whether depressive symptoms were occurring in parents caring for a child with chronic conditions is the impact that it has been said to have on the child. In a review by Rodenburg, maternal depression was said to contribute to the likelihood of a child developing psychopathology.(Rodenburg et al. 2005) Chiou et al found that higher parental depression was associated with children diagnosed with epilepsy having a poorer self-concept.(Chiou & Hsieh 2008a) A study involving 51 mothers showed that increased maternal depression was significantly associated with child maladjustment when their child was diagnosed with a conduct disorder. (Dumas, Gibson & Albin 1989)Shore et al also supported the notion that maternal depression has a wide impact when the diagnosis is epilepsy, by stating that depressive symptoms caused a disruption in family routines and day-to-day life.(Shore et al. 2002)

Carer Locus of Control

A locus of control refers to the extent that an individual believes that their actions influence events.(Perrin & Shapiro 1985) Rotter first proposed that there are two variations in a person's locus of control, that they are either external or internal.(Rotter 1990) External locus of control is the degree that an outcome is viewed a being 'a function of chance, luck or fate, is under the control of powerful others, or is simply unpredictable'.(Rotter 1990) Internal is the opposite of this notion, which an event occurs to individuals as a 'contingent on their own behaviour and characteristics'.(Rotter 1990)

The interest in the belief that individuals have different locus of control is due to the concept that it is a mediator between stress and wellbeing.(Glenn et al. 2009; Parkes 1984; Siman-Tov & Kaniel 2011)

Studies that have examined the relationship between stress and locus of control have suggested that an external locus of control is positively correlated with increased parental stress experienced when a child has been diagnosed with a chronic condition. (Glenn et al. 2009; Siman-Tov & Kaniel 2011) Goldbeck also echoed these sentiments by describing external locus of control as being a maladaptive technique due to the passive patient behaviour that the beliefs generate.(Goldbeck & Bundschuh 2007)

A study by Perrin et al compared locus of control between mothers of children with no health complaints, the control group, to mothers of children with seizures. No group sizes were given. Perrin et al demonstrated that mothers of children with seizures obtained higher scores for beliefs in the subgroup 'powerful others' and slightly lower scores in the subgroup 'internal' than the normal population. Overall it was said that mothers exhibited a more external locus of control.(Perrin & Shapiro 1985) Another study of 187 parents of children with different health conditions, including epilepsy, found that maternal locus of control and a child's intelligence level predicted child adjustment, with internal locus of control producing a strong positive relationship with adjustment.(Perrin, Ayoub & Willett 1993)

Carer Coping

Austin et al described coping as 'action strategies related to family relationships, parenting and supporting the child's successful adjustment'. (Austin & Caplan 2007)

Duffy stated in her review of parental coping and childhood epilepsy, that it was essential for parents to maintain their coping strategies, as by doing so, parental wellbeing is preserved and therefore family wellbeing will be too.(Duffy 2011) Mu et al supported this notion, stating that in the study of 316 mothers whose child had been diagnosed with epilepsy, efficient coping patterns were found to maintain family coherence and unity.(Mu, Kuo & Chang 2005)

Carver et al stated that coping is context-dependent, that the way in which people cope is derived from the nature of the stress and the interactions between the stressors and the environment.(Carver, Scheier & Weintraub 1989) However, different studies have indicated that specific types of coping strategies are associated with more stress.

In Rodenburg's review of parents whose child has epilepsy, it was suggested that a general lack of parenting coping resources might cause increased parenting stress.(Rodenburg et al. 2005) Rodenburg et al identified in their study of 91 parents of children with epilepsy, that particular types of coping strategies either served to increase or decrease parenting stress. Problem focused coping strategies decreased parenting stress, whilst emotional focused coping strategies were a significant contributor to parental stress.(Rodenburg et al. 2007)

Mitchell et al followed 119 families presenting with a child having seizures over at least 6 months and found that families who used more medical guidance and greater

physician contact, acted as a form of instrumental support. This resulted in a reduction of parental stress and a greater adherence to medication.(Mitchell, Scheier & Baker 2000) This may highlight the different coping strategies employed when parents feel that they are under a lot of stress, that those experiencing significant stress will seek medical guidance and physician support as a form of instrumental coping mechanism.

2.9.3 The Effect of a Diagnosis of Epilepsy on a Child

Quality of Life of the Child

Epilepsy is associated with reduced quality of life.(Devinsky et al. 1999; Hoare 1993; Taylor et al. 2011a; Taylor et al. 2011b) Health-related quality of life (HRQOL) is described as the 'individual's self perception of their physical, mental and social wellbeing.'(Devinsky et al. 1999) Devinksy et al found from a study involving 197 adolescents diagnosed with epilepsy, that poorer HRQOL was associated with older age, more severe epilepsy and lower socio-economic status.(Devinsky et al. 1999) Modi et al compared HRQOL between those with newly diagnosed epilepsy and those who had presented with a single seizure. Using data from 109 children, it showed that their HRQOL measure was significantly lower than normative data, but the difference between the groups was not significant.(Modi et al. 2009) As part of the SANAD trial, HRQOL measures were also undertaken, comparing children with newly diagnosed epilepsy to those with an established diagnosis of asthma. They found that those newly diagnosed with epilepsy reported significantly overall poorer quality of life across physical, emotional, self-esteem, friend and school domains. They suggested that the lower quality of life in multiple domains might be specific to epilepsy. However they also identified that the duration of diagnosis for the two comparison conditions were different, implying that a new diagnosis has not allowed an individual to habituate to the change in their health status, resulting in lower HRQOL scores.(Taylor et al. 2011a) The finding of lower HRQOL scores in children diagnosed with epilepsy, agrees with research that depression is reported regularly with an estimated prevalence rate of 23-26%.(Austin et al. 2010; Cushner-Weinstein et al. 2008; Dunn, Austin & Huster 1999) Loney et al additionally stated that those presenting with first seizure were also experiencing depressive symptoms. Significantly higher depression scores

were found from the 22 children with first seizure compared to 42 children in a control group with non-neurological presenting complaints ($p<0.05$).(Loney et al. 2008)

Child self-esteem

Self-concept is cognitive appraisal conducted by you about yourself, with the individual evaluation of yourself being self-esteem.(Hoare & Mann 1994) A positive self-concept has been said to be central to adaptive functioning in an individual.(Butler & Gasson 2005) Conversely, a negative self esteem and image is related to negative health effects such as depression.(Butler & Gasson 2005)

It is thought that low self-esteem is more likely to occur in people with chronic conditions due to the limitations that are placed on their physical and social functioning.(Vilhjalmsson 1998; Wallander & Varni 1998) Epilepsy has been associated with low self esteem in children.(Stafstrom & Havlena 2003) Stafstrom et al asked 105 children diagnosed with epilepsy to draw a picture of what it was like to have epilepsy. From their findings, they concluded that across all syndromes and ages there was evidence of impaired self-concept and low self-esteem.(Stafstrom & Havlena 2003)

Austin et al found in a longitudinal study of 135 children that high parenting stress was associated with a decrease in child self-esteem and a decline in processing skills. Poor self-esteem was also associated with poorer cognitive abilities.(Austin et al. 2010) A cross-sectional study comparing children diagnosed with epilepsy (n=62) to those with diabetes (n=91) found that the epilepsy group reported lower self esteem and higher behavioural disturbances.(Hoare & Mann 1994)

Child behaviour problems

Children with chronic conditions, especially neurological conditions such as epilepsy, have been shown to be at greater risk for behavioural problems.(Hoare & Mann 1994) A child's behavioural problems have been posited as a common stressor for parents who have a child with a chronic condition.(Buelow et al. 2006)

Hoare et al conducted semi-structured interviews and questionnaires with 103 carers of children with epilepsy and found that family stress and negative parenting attitudes were associated with behavioural problems in children with epilepsy.(Hoare & Kerley 1991)

Wirrell et al found that 80 mothers of children with intractable epilepsy reported significantly higher stress levels than normative data, with a significant correlation between stress and the externalising and total problems identified in their child via the Child Behaviour Checklist.(Wirrell et al. 2008)

A study of 51 children with epilepsy and their parents by Pianta et al, found that increased parental stress and poorer parent-child interactions, were associated with more child behaviour problems.(Pianta & Lothman 1994)

2.9.4 The Effect on the Family when a Child has been Diagnosed with Epilepsy

Family functioning

The impact of epilepsy upon the family functioning was identified in the semi-structured interviews conducted by Hoare et al. Using the data from 35 mothers of children with epilepsy compared to families who did not have a child with epilepsy, it was found that epilepsy restricted family activities and increased the required level of child care, producing significantly higher stress scores when compared to comparison families.(Hoare & Kerley 1991) It is not just the direct impact that epilepsy has upon family life that is of interest, but how studies have identified that the family environment serves as a moderator towards the effects that epilepsy has.(Austin et al. 2004a; Baum et al. 2007; Mitchell, Scheier & Baker 2000)

The family environment is important to consider as the characteristics of the family as a unit have been attributed to contributing to the psychological distress felt by it's members as well as acting as a preventer.(Austin et al. 2004a; Holmes & Deb 2003)

Rodenburg found from the study of 91 parents of children with epilepsy that a cohesive family environment resulted in a reduction of parental stress.(Rodenburg et al. 2007)

Mitchell et al longitudinal study of 119 children with epilepsy, also found that the family environment was associated with the amount of parental stress being felt, with an organised family environment causing less parental stress.(Mitchell, Scheier & Baker 2000)

Thornton et al conducted a cross-sectional study involving 82 cognitively normal children who were diagnosed with epilepsy and compared them to a similarly aged sibling without a diagnosis of epilepsy. They found that family functioning for these

families was the same as a normal population but scored higher in the area for involvement. This study felt that it indicated that epilepsy had a positive effect on the family environment, as the family members were more empathetic towards each other with supportive interactions being displayed. The did find however that parents scored significantly poorer than normal values in their ability to adapt to the new parenting role demands that a diagnosis of epilepsy produces.(Thornton et al. 2008)

A poorer unity between all family members has been associated with increased stress, with Holmes et al stating that psychological distress could be described as being contagious when the family structure is not cohesive.(Holmes & Deb 2003) McCusker et al conducted a cross-sectional study involving 48 families who have a child with intractable epilepsy and found that the family environment was the second most important element in predicting a child's behavioural and adjustment difficulties.(McCusker et al. 2002) The inverse relationship between family cohesion and behavioural problems as found by McCusker et al, was a finding found to be reiterated when a review of epilepsy was conducted to examine the relationship between family factors and stress.(Rodenburg et al. 2005)

Austin et al found in a longitudinal study of 224 children with new onset epilepsy, lower family functioning along with poor parental confidence was associated with more behavioural problems.(Austin et al. 2004a) Austin has also identified that a positive family environment acted as a protective factor for a child's self-esteem when those children had a cognitive decline.(Austin et al. 2010)

Support

Social support has been shown to act as a moderator towards daily and also chronic stress.(Thompson & Upton 1992) Rodenburg et al found from the study of 91 parents that higher levels of social support produced lower parenting stress.(Rodenburg et al. 2007)

Thompson et al found that primary carers to children with chronic epilepsy felt they had inadequate support with most received from the family. Within this study, 6 of the 44 caregivers felt they got considerable support from external services. The general lack of support was seen to be significantly associated with poor emotional adjustment.(Thompson & Upton 1992) Eiser et al found that mothers of children with epilepsy reported social support/information to become less helpful over time since

their child was diagnosed, suggested to occur due to support being withdrawn over time.(Eiser et al. 1992) Austin et al also reported in a study involving children with new onset epilepsy, that many parents felt that the social support being received was insufficient and that they needed additional help specifically from healthcare providers.(Austin 1996)

An alternative way to assess support is to evaluate the needs that carers express. The importance of meeting needs is the potential that it has in reducing the negative aspects of the condition, such as fear and stigma, and by increasing a sense of control.(Couldridge, Kendall & March 2001) A systematic review on needs revealed that patients and carers felt that the provision of information about epilepsy was a continuing and unmet need.(Couldridge, Kendall & March 2001) McNelis et al found that there was an association between unmet information needs and increased anxiety. (McNelis et al. 2007) This highlights the powerful impact that ensuring patients have the information they want as a means of increasing their quality of life. Shore et al also found in a longitudinal study over 24 months that a high proportion of the 143 families involved within the study expressed unmet needs despite the seizures being well controlled. The expressed needs regarded information on epilepsy and the available treatment as well as more support provision.(Shore et al. 2009) The authors hypothesised that the continuing unmet needs may be due to epilepsy producing different needs at different stages, only becoming apparent when the parents learn and experience more of the condition.

From the literature reviewed within chapter two, it is clear that there have been many studies examining the impact on the child and their family when they are diagnosed with epilepsy. It does appear that there are fewer studies that have examined the impact when the child is newly diagnosed with epilepsy.

A critical appraisal of the papers that have examined parenting stress and newly diagnosed epilepsy in their child will be performed. This will help firstly to identify how many studies have been conducted in parenting stress and newly diagnosed paediatric epilepsy.

The critical appraisal was to rigorously assess studies that are examining a similar group of participants as used within this present study. The aim was to examine how the previous studies have been conducted and their findings. The findings revealed by the critical appraisal can then be compared to this present study's findings. It will then help to identify the methods used by these studies and their findings.

3.1 Aim

To review studies examining the factors that affect stress in parents of children under 18 years who have been given a new diagnosis of epilepsy.

3.2 Methods

3.2.1 Inclusion criteria:

1. Research papers using qualitative or quantitative observational research methods to explore factors affecting parental stress when their child has been given a new diagnosis of epilepsy
2. Children aged 0-18 years who have been diagnosed with epilepsy
3. Journal papers published in English up to July 2011 (i.e. excludes dissertations, books, book chapters and reviews)

3.2.2 Exclusion criteria:

1. Studies that did not state duration of epilepsy diagnosis to be under two year in a paediatric population (0-18 years)

2. Review articles

3.2.3 Search strategy

In order to increase the probability of identifying all relevant studies, a variety of different databases were searched. The databases used to find the most relevant papers were Medline (1948-current), Scopus (1823-current), PsychInfo, and CINAHL.
The keywords used within the search were "parent", "stress" and "epilepsy". The three keywords were combined using the Boolean operator AND so to narrow the search and to identify the relevant studies.
In order to identify the full number of relevant studies, it was accounted that synonyms of the keywords may be used. This was achieved by using major subheading (MESH) when searching through Medline and by accounting for alternative spellings for "parent" by applying an asterix. The search was performed in July 2011. The full search strategy can be seen in Appendix 1.

3.2.4 Data Extracted from Each Paper

Each abstract was read for every paper that appeared within the search performed in order to assess its eligibility. If the paper appeared to be relevant from this, the full text was assessed to see if it meets the eligibility criteria that had been set.

Once the eligibility of the paper had been clarified, each study had the following data extracted from it:

1. Duration of epilepsy diagnosis
2. All factors investigated for being associated with parental stress
3. Study features: year of publication, sample size, study design, ages of children
4. Seizure characteristics: Seizure frequency, seizure type.

3.2.5 Evaluation of Study Quality

The following articles were appraised using either the proforma developed by Critical Appraisal Skills Programme (CASP) Appraisal Tool for qualitative research (CASP 2006b) or an adapted version of the Critical Appraisal Skill Programme for case-control research (CASP 2006a) so to account for different types of observational study designs. A table was then created using the headings from the proforma and each paper

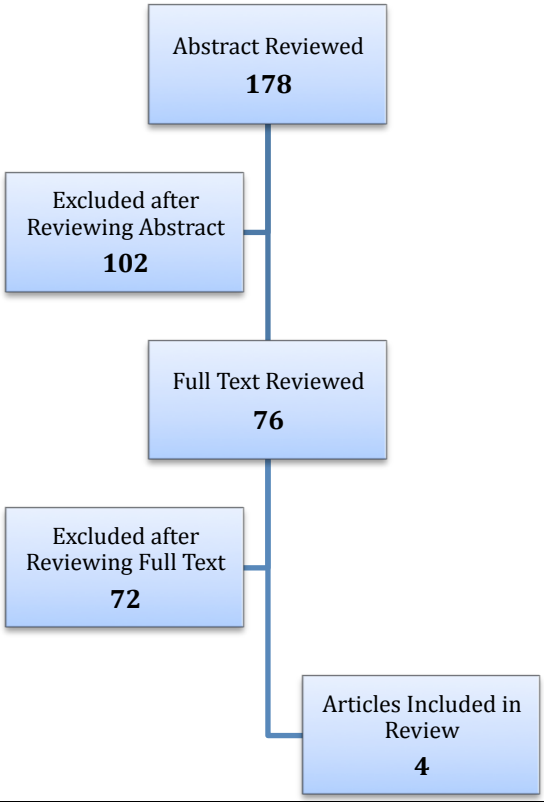
was individually assessed for the quality of the research and as a means of summarising their findings.

3.3 Results

3.3.1 Study Selection

The search performed produced 178 potentially eligible studies. From the 178 articles, 102 were excluded after reading their abstract to identify 76 papers that should be retrieved in full. From the 76 papers, 72 were excluded due to not fulfilling the eligibility criteria that had been previously set. This resulted in 4 articles that were then to be critically appraised. A flowchart diagrammatising the selection procedure of the articles to be critically appraised can be seen in Figure 7.

Figure 7: Flow Diagram of Identifying Relevant Articles



3.3.2 Description of the Articles

From the search, four articles were identified as being relevant. The duration of epilepsy diagnosis for the four papers ranged from 0-18 months, with three of the studies conducted within 6 months of a child being diagnosed with epilepsy.
The study design varied with three of the studies being of an observational design and one being a qualitative study design. The three observational studies used health-related stress questionnaires, being either Family Inventory of Life Events and Changes or the Parent Stress Index. Only one study used an additional epilepsy specific scale of parenting stress.
Of the population who participated in the study, two were solely recruiting mothers of children with new-onset epilepsy with the other two studies recruiting both the mother and father to participate.
The age of the children who had been diagnosed with epilepsy was different for each study, with the largest age range being 2-12 years. Three of the studies had an upper limit of 12 years old. Only one paper specifically stated an objective of examining parental stress whilst the other three papers assessed for parental stress although the main variable of interest was another factor. A summary of the four papers to be critically appraised can be seen in Table 4. The process of the critical appraisal for each study can be seen from Tables 5-8.

Table 4: Summary of the Data Extracted from Each Article

Article	Author	Year	Duration of epilepsy diagnosis	Sample size	Study design	Age of children	Population involved	Objective	Outcome
Do depressive symptoms affect mothers' reports of child outcomes in children with new-onset epilepsy?(Ferro et al. 2010)	Ferro MA et al.	2010	Completed at diagnosis	339	1 time point. Cross-sectional analysis of baseline. Questionnaires	4-12 years	Mothers of CWE	Assess for psychological distress and the effect it has on their report of a child's quality of life.	Families demonstrated that they were functioning well with good resources and low stress. Did not find that maternal psychological distress affected their reports on their child's quality of life.
The Impact of a New Pediatric Epilepsy Diagnosis on Parents: Parenting Stress and Activity Patterns(Modi 2009)	Modi AC	2009	1 month	59: 30 epilepsy; 29 control	1 time point Case-control matched study Questionnaires and 2 daily phone diaries for 24 hours.	2-12 years	Parents of CWE, parents without CWE	Compare parenting stress and activity patterns of parents with CWE to those without epilepsy	No significant difference in stress scores between two groups. Significant correlation between parental stress to CWE and recreation time at home. Parents of CWE spent more time in medical care and less recreation time.
Transition experience of parents caring for children with epilepsy: A phenomenological study(Mu 2008b)	Mu PF	2006	18 months	18 (10 couples)	1 time point Colaizzi's in-depth interview	3-7 years	Parents of CWE	Investigate parental perspective of epilepsy diagnosis in the first 18 months	Parents were emotionally traumatized and physically exhausted following diagnosis, stigmatized with 'loss' of healthy child. Perception of medically controlled epilepsy reduced stress. Instrumental support reduced stress.
Adaptive functioning in children with seizures: Impact of maternal anxiety about epilepsy(Chapieski et al. 2005)	Chapeski L et al.	2005	6 months	56	2 time points over 1 year. Longitudinal study. Questionnaires and semi-structures interview.	6-12 years	Mothers of CWE	Investigate impact of maternal anxiety on parental patterns and child's adaptive functioning	High levels of maternal anxiety were predicted with more family stress and fewer coping resources. Higher anxiety and stress was associated with lower levels of adaptive functioning. Evidence that stress may aggravate anxiety.

3.3.3 Critically Appraised Articles

Table 5: Do Depressive Symptoms Affect Mothers' Reports of Child Outcomes in Children with New-Onset Epilepsy? (Ferro et al. 2010)

Criteria	Success in addressing the criteria
Screening questions	
Was there a clear statement of the aims of the research?	To test whether elevated levels of depressive symptoms in mothers affected their report of health-related quality of life in their child with new-onset epilepsy.
Is the study methodology appropriate?	Cross-sectional observation using 6 questionnaires to assess family functioning, family resources, stress, maternal health and child wellbeing.
Detailed questions	
Was the population studied appropriate and recruited in an acceptable way?	Cases were 339 English-speaking primary maternal caregivers of children aged 4-12 years. Recruited by paediatric neurologists at different neurological clinics across Canada. All are part of a prospective cohort study.
Were the measurements used appropriate?	Objective measurements were used through validated questionnaires. The depression questionnaire actually assesses psychological distress rather than a tool to diagnose depression.
Were confounding factors accounted for?	Confounding factors were controlled by using questionnaires that assessed mothers' age, education, employment, family functioning, social resources and stress.
What are the results of the study?	That families were functioning well, had low stress and adequate resources. No evidence found that maternal reports on child outcomes are affected by depressive symptoms.
How precise are the results?	Unadjusted and adjusted regression analyses were run to assess the effect of controlling or not controlling confounding factors. Bonferroni correction was applied to account for multiple testing.
Were all the important outcomes considered?	Seizure characteristics were not presented.
Can the results be applied to the local population?	External validity may be limited due to all cases recruited from paediatric neurological clinics and may not represent all families.
Do the results of this study agree with other available evidence?	Evidence examining affect of maternal health on their reports of child's behaviour

Table 6: The Impact of a New Pediatric Epilepsy Diagnosis on Parents: Parenting Stress and Activity Patterns (Modi 2009)

Criteria	Success in addressing the criteria
Screening questions	
Was there a clear statement of the aims of the research?	To compare parenting stress and activity patterns in parents of children with new-onset epilepsy and parents of children without epilepsy.
Is the study methodology appropriate?	Case-control study using validated questionnaires to measure parenting stress and seizure-specific stress. Two retrospective 24-hour daily phone diaries were used to assess parents' opinion on their activities.
Detailed questions	
Was the population studied appropriate and recruited in an acceptable way?	Clear set criteria for recruiting cases: Parents of children presenting to paediatric clinics in the Midwest with a new diagnosis of epilepsy that requires treatment. Controls were recruited mainly from hospital wide email. Matched for gender, race and +/- one year of age. One fewer control than case.
Were the measurements used appropriate?	All questionnaires have been previously validated. Daily phone diary demonstrated >90% interrater reliability with open-ended questions to prompt reconstruction of daily activity. Controls and cases completed the same methods. Blinding of researchers was not undertaken.
Were confounding factors accounted for?	States that confounders were controlled but does not describe what they were.
What are the results of the study?	No significant differences between cases and controls for parenting stress. Higher proportion of parents of children with epilepsy spent more time in medical care and less recreation time at home. Significant correlation between epilepsy-specific parenting stress and recreation time at home.
How precise are the results?	Small sample size with 94% agreeing to take part. No significant differences found between cases and controls for stress. Significant activity differences between cases and controls with $p<0.05$. Epilepsy-stress correlated to recreation time at home was $r=0.47$, $p<0.01$.
Were all the important outcomes considered?	States that it is likely that nighttime waking had occurred in their sample of parents but did not assess for this. Social support and resources were not considered and nor were epilepsy specific variables.
Can the results be applied to the local population?	The sample population is the same as the population to be enrolled in the study presented by the thesis, suggesting that the same results may be found.
Do the results of this study agree with other available evidence?	Parenting stress was reported to be present in only 7% of parents, equating to 2 of the 30 parents involved in the study. This is a lot less than previous evidence.

Table 7: Transition Experience of Parents Caring for Children with Epilepsy: A Phenomenological Study (Mu 2008b)

Criteria	Success in addressing the criteria
Screening questions	
Was there a clear statement of the aims of the research?	To investigate how parents have found having a child being diagnosed with epilepsy by reflecting on their experience. Aim to understand further parental reactions.
Is the qualitative methodology appropriate?	Colaizzi's phenomenological approach was used with in-depth interviews of parents. The inter-subjective allowed parents to express their concerns and difficulties of having a child being diagnosed with epilepsy.
Detailed questions	
Was the research design appropriate to address the aims of the research?	States that an open attitude and imaginative techniques were used to investigate the meanings of individual experiences.
Was the recruitment strategy appropriate to the aims of the research?	Separate pilot study had been performed prior to study. Purposive sampling was employed from two medical centers in Taiwan to enroll 10 parent couples with two fathers who did not take part. Only enrolled parents who were intently interested in understanding the health-illness process.
Were the data collected in a way that addressed the research issue?	Open-question interviews at unspecified setting that was recorded and transcribed verbatim. Simultaneous information obtained from both parents to obtain family experience from two perspectives. States the starting questions and describes the use of facilitative techniques. States data was analysed for thematic content and if repeated, data saturation had been reached.
Has the relationship between the researcher and the participants been adequately considered?	One researcher performed all interviews and acknowledges interviewer bias by stating that the researcher set aside personal bias and assumptions and a research journal was used to record the researcher's reactions. Used guidelines suggested by Lincoln and Guba for methodological rigor.
Have ethical issues been taken into consideration?	The Hospital Human Investigation Committee at each of the two medical centers gave ethical approval. States that the participants' were informed at the time of interview that they had the right of anonymity, confidentiality and study withdrawal at any time.
Was the data analysis sufficiently rigorous?	Analysed manually using Colaizzi's method that integrates both destructured and restructured analysis to suggest three major domains. Three individuals conducted the analysis to control its consistency, stability and reproducibility.
Is there a clear statement of findings?	Three major domains identified: parental psychological reactions, coping patterns and family resources. Findings agree with family stress theory and other research examining the health-illness transition.
How valuable is the research?	Identifies that the stigma experienced reflects Chinese culture and may not be reproducible to other populations. Also states that the characteristics represent families who were willing to share their experience. Suggests future work to examine transition when child has been diagnosed with epilepsy and another condition. States that family centered management for parents would provide them better support and assist in a child's development.

Table 8: Adaptive functioning in children with seizures: Impact of maternal anxiety about epilepsy (Chapieski et al. 2005)

Criteria	Success in addressing the criteria
Screening questions	
Was there a clear statement of the aims of the research?	Stated that several maternal factors were going to be explored to assess maternal anxiety, parenting styles and a child's adaptive behaviour in relation to their child being newly diagnosed with epilepsy.
Is the study methodology appropriate?	Longitudinal study design to examine the relationships over time. Used validated questionnaire instruments to assess relationships. Appropriate measures used to examine anxiety, stress, coping, maternal protectiveness and child behaviour.
Detailed questions	
Was the population studied appropriate and recruited in an acceptable way?	56 mothers and one of their children were 80% recruited from schools and 20% recruited from paediatric neurologists in Texas. Children had no other co-morbidity or behaviour problem other than newly diagnosed epilepsy. No method stated for recruiting participants. Two data collection points at 0 and 1 year, attrition rate of 25%.
Were the measurements used appropriate?	Objective measures of previously validated questionnaires. Semi-structured interview was performed without any mention of the qualitative results. Anxiety measure had not been used on this study population before.
Were confounding factors accounted for?	Gender, race, socio-economic status, seizure characteristics, AED use were all accounted for. Multiple regression analysis was used to assess relative contribution of these factors.
What are the results of the study?	Higher maternal anxiety was associated with lower socio-economic status and more family stress and fewer resources when initially presenting with child. High maternal anxiety and family stress were associated with lower levels of adaptive functioning at first visit. This relationship was non-significant at second visit.
How precise are the results?	Multiple regression analysis between anxiety and stress at first visit was 0.06, $p<0.04$. 25% of families did not return for second visit. No mention as to how analysis accounted for the loss of numbers. Anxiety measure had not been previously used in this population.
Were all the important outcomes considered?	Seizure frequency was not assessed due to the study population experiencing fewer seizures. Family resources were not considered.
Can the results be applied to the local population?	Population was from only the mother perspective. The study group represented children who had few seizures. Population was obtained from both community and hospital settings, which is different to the thesis study.
Do the results of this study agree with other available evidence?	Attempts to use study findings as a means of explaining previous research. It is not apparent from study whether the findings are new or agree with any previous research.

3.4 Discussion

This review identified four papers that reported to measure stress in parents of children with newly diagnosed epilepsy. As the interest of the review was to examine newly diagnosed epilepsy, this reduced the number of studies available to analyse. It was found that surprisingly few studies specified the epilepsy diagnosis duration with only these four papers controlling a short duration of epilepsy diagnosis. Of the four papers reviewed, only the study by Modi et al had the specific objective to examine parental stress. The remaining three studies reported upon parental stress in their specified population with the aim being to understand the relationship of a different variable. Chapieski et al examined parental stress in order to delineate maternal anxiety when a child is newly diagnosed with epilepsy. Ferro et al measured parental stress to understand the relationship between maternal depression and their report of their child's quality of life, whilst Mu et al reviewed parental experience of having a child newly diagnosed with epilepsy and found that parental stress was being reported.

The location of the studies varied, with two taken place in the United States of America(Chapieski et al. 2005; Modi 2009), one in Canada (Ferro et al. 2010) and another in Taiwan(Mu 2008b). Only the study in Taiwan reported upon the effect that a cultural belief of epilepsy has on parental stress. The remaining three studies measured the demographic characteristics of the studied population but without assessing cultural origins.

As the study to be undertaken and presented within this thesis is based within the United Kingdom, it may be fair to assume that the stigmatisation felt by those within Taiwan, reflects the Chinese culture of epilepsy being a 'sudden craziness' and may not be a view reflected within western culture. Mu et al found that parents who shared the Chinese cultural belief or thought others would perceive it as 'a sudden craziness', caused an exacerbation of parental stress. The role of stigma and the effect that it has upon the child who is diagnosed with epilepsy as well as their parents has been reported in western cultures.(Austin et al. 2004b; de Boer 2010) However, it could be debated, as the present study is recruiting carers from a western culture, that stigma will not affect parental stress as greatly as the parents recruited by Mu.

Parental stress was measured in different ways, with Ferro et al and Chapieski et al using the Family Inventory of Life Events and Changes (FILE) measurement tool and Modi et al using the Parenting Stress Index (PSI) and Family Stress Scale-Seizure

measurement tools. Two studies reported that parents were experiencing low stress. (Ferro et al. 2010) (Modi 2009) It was additionally identified in one paper that there was no significant difference in stress scores between parents of children with newly diagnosed epilepsy and parents whose child has no medical complaints.(Modi 2009) Only one paper specified that parents were likely to feel stressed. (Mu 2008b) One paper involved two time points over 1 year, finding that the mean stress scores of the parents remained similar (T1=11.56, T2=11.58).(Chapieski et al. 2005) Unfortunately there was no attempt within the paper to suggest a reason as to why the stress scores have remained unchanged.

The relationship between parental stress and other variables was examined by three of the studies. Modi et al examined the relationship between parental stress and their activity patterns, finding a trend of parents to children with epilepsy spent more time in medical care and less in recreation than the control group, as well as identifying a significant correlation between epilepsy-specific stress and amount of recreation time spent at home ($r=0.47$, $p<0.01$). They suggested that this was due to lack of social support or providing enough information to help parents feel they can manage.(Modi 2009) However it should be noted that the sample size for comparing epilepsy-specific stress and activities was 30 as it only involved the parents who has a child with newly diagnosed epilepsy.

Mu et al found from the interviews conducted that increased social support, increased information and increased family functioning helped to reduce the amount of parental stress.(Mu 2008b) Chapieksi et al also identified the role of support, suggesting that mothers with increased social support were less anxious. It was further found that increased parental stress was associated with maternal anxiety, producing a correlation of $r=0.38$, $p<0.01$. A multiple regression analysis found that anxiety predicted increased stress and reducing coping ability when a child was initially diagnosed with epilepsy, $p<0.04$. They additionally found that when a child is initially diagnosed with epilepsy, increased parental stress and increased anxiety both were associated with a child less likely to adapt to the diagnosis.(Chapieski et al. 2005) Ferro et al did not find that parental stress was related to maternal depressive symptoms affecting a mother's view of their child's quality of life.(Ferro et al. 2010) The role of parental stress affecting parent health was suggested to contribute to maternal anxiety by Chapieski et al.(Chapieski et al. 2005) Modi et al additionally implied that parental anxiety may be

the more pressing issue than feeling stressed when a child is initially diagnosed, however anxiety was not formally measured within the study.(Modi 2009)

The different papers considered epilepsy-specific characteristics, with Modi et al finding no significant differences in stress scores between parents of children that had had a seizure since diagnosis and parents whose child had not had a seizure. Mu et al stated that a primary concern of parents was that they did not know when the next seizure would occur, echoing statements identified in previous literature that the uncertainty of epilepsy is a principle cause of parental stress. (Murray 1993; Nolan, Camfield & Camfield 2006) Mu et al suggested support for this and found that well controlled seizures reduced parental stress. (Mu 2008b)

3.5 Conclusion

From reviewing these studies, it is apparent that there are differences in whether parental stress is experienced when a child is diagnosed with epilepsy. The only study that stated that parents were stressed from their child being diagnosed with epilepsy was the interview-based study. (Mu 2008b) The reason for this could have been that only 18 individuals were involved in the study and were recruited if they had an interest in the health-illness transition. This does suggest that there was a selection bias by only recruiting parents who felt that there had been a large alteration from having a healthy child to one who is now diagnosed with a chronic condition. It also studied parents whose child had been diagnosed the longest, increasing the probability of a parent having experienced an epilepsy-specific stressful event. However, the effect of time on parental stress was examined by Chapieski and found that over a one-year period, mean parenting stress levels remained the same. (Chapieski et al. 2005)

The examined studies appears to find that the available resources to parents, acts as a mediator to parental stress. Mu et al identified that instrumental and social support were direct mediators of parental stress. Chapieski identified a relationship between maternal anxiety, stress and coping resources and Modi et al suggested that support was a mediator between stress and family activities undertaken, although this was not formally assessed.

Although it cannot be certain if parents experience stress when their child is newly diagnosed with epilepsy, it appears that stress has the potential to affect family activities and family member's health and that using resources can act as a buffer of

stress. There were limited quantifiable data presented that explored the mediating factor of parental stress. It was suggested that resources and family functioning were important in moderating parental stress, however further work is needed to explore if there is a relationship that can be quantified. The model presented by Chapieski et al suggested that increased stress and reducing coping predicts anxiety. It would be useful to examine if there is a relationship between stress and coping and further whether specific coping styles are more useful at reducing parental stress. It would therefore be beneficial to explore whether parental stress is associated with family variables such as support, functioning and ability to cope as well as determining if there is a direct relationship between carer health and stress. Therefore, the study presented within Chapter Five will focus upon examining the effects of mediating factors on parental stress.

Chapter Four

Interim Analysis of the Correlates of Stress Adjustment in Carers of Children with Newly Diagnosed Epilepsy:

Stress and its Covariants in Carers of Children with a Possible Diagnosis of Epilepsy.

4.1 Introduction

The following sections of the thesis will be presenting the preliminary analysis of the data obtained for the main study 'Stress adjustment in carers of children with newly diagnosed epilepsy'. The data were collected over a 12-month period, with all data representing the initial presentation to an outpatients department with a potential diagnosis of epilepsy being suspected. The main study aims to enrol 200 patients, all who have presented with a suspected diagnosis of epilepsy. This is in order to create two groups of patients; one who later are diagnosed with epilepsy and the other group to represent those who do not get diagnosed with epilepsy.

The future aim of the main study is to perform Structural Equation Modelling (SEM) as a means of assessing the effect that variables have on the stress being experienced by carers. This main study aims to examine complex relationships between stress and the variables, within two different populations groups at two separate time points. A quantitative methodology was therefore employed, as the results would allow the ease of examining these different relationships via SEM.

As stated, the results and analysis will be presenting a cross-sectional analysis of all those enrolled into the study so far, with all children only having the potential diagnosis of epilepsy.

4.2 Objectives

4.2.1 Aims

This thesis is presenting an interim analysis of a 3-year study into correlates of stress adjustment in carers of children with newly diagnosed epilepsy. The aims of this thesis are as follows:

1. To explore whether carers are experiencing stress when their child is initially presenting with a potential diagnosis of epilepsy at an outpatient department.

2. To assess if the response to the different variables being measured varies with each family enrolled in the study.

3. To assess if there are any associations between specific variables as reported by the carer and their stress scores.

4.2.2 Hypothesis

The different hypothesis for each aim is as follows:

1. Carers will be experiencing stress when they are initially presenting in an outpatients department.

2. There will be a difference in scores given by the carers and the children that complete each questionnaire, serving to measure a specific variable.

3. There will be some variables, as measured by questionnaires completed by the carer, which will be associated with carer stress.

4.3 Methodology

4.3.1 Administrative Organisation

This single centre study was carried out primarily in the Neurology Outpatients Department but also within the General Outpatients Department, both situated at Alder Hey Children's Hospital. All participants were approached, recruited and seen in an outpatient department. Funding was provided by the Alder Hey Charitable Trust fund, specifically from the Neuro-disability Charitable Trust Fund.

4.3.2 Ethical Approval

Ethical approval was granted in full from both the NHS Research and Development Offices and the Northwest 3 Research Ethics Committee-Liverpool East.

The Research Ethics Committee reference number is 09/H1002/91.

Full ethical approval for the current protocol described in this study was granted on the 22nd September 2010.

4.3.3 Study Population

The inclusion criteria for this study was children of both genders aged 0-16 years and who were being referred to an outpatient department within Alder Hey Hospital, Liverpool, with epilepsy being a potential cause for the symptoms they were experiencing.

4.3.4 Principal Inclusion Criteria

The principal inclusion criterion was:

1. The caregiver must have a child who was being referred due to experiencing seizures with epilepsy being a suggested diagnosis.
2. Child must be between the ages of 0-16 years
3. Child must be a registered patient at Alder Hey Children's Hospital
4. The caregiver must have a reasonable understanding of the study, understand what they will have to do for the study and be competent enough to refuse participation.

4.3.5 Principal Exclusion Criteria

The principal exclusion criterion was:

1. A child with a prior or established diagnosis of epilepsy
2. A child diagnosed with another relapsing-remitting condition that required regular medication.

4.3.6 Justification for the Criteria

Age

Initially the protocol stated that children would be recruited if aged 6-16 years. However, it was noticed that a substantial number of potential recruits were being missed due to the lower age limit in place.

Under approval from the local ethics research committee, the age limit was lowered to newborn children with only the carer to complete the questionnaires. As the primary aim of the study was to assess the stress experienced by the main carer, it was felt appropriate to reduce the age limit for potential recruits, despite the child not being able to partake if they were below the age of 6 years. The child is not able to complete the questionnaires if they are below 6 years as the questionnaires completed by the children are only valid from 6 years and above.

It is acknowledged that this is a limitation of the study as there is loss of data for these children.

The upper age limit was created due to the long-term plan of the study; that all carers and children recruited will complete the questionnaires for the second time between 6 months to 1 year after initial completion. The age limit of 16 would therefore help to ensure that fewer patients are lost to follow-up, as a transfer to adult services should not have yet occurred.

Previous or Established Diagnosis of Epilepsy

An established diagnosis of epilepsy was an exclusion criterion in order to assess the impact of the diagnosis by comparing scores before and after a diagnosis is given. The rationale for this is that the study is aiming in the future to target families early, at initial presentation, and to provide them with support so to avoid parental stress. By comparing scores before and after a diagnosis of epilepsy, it will provide guidance as to type of support that is needed.

Previous diagnosis of epilepsy was excluded, as the aim of the study is to assess the impact that a suggested diagnosis of epilepsy has upon the family and the child. The reason for this criterion was that if a previous diagnosis of epilepsy had already been experienced, the situation would be familiar to the family. This is to avoid recruiting participants who may have already adjusted to the stressor. It has been shown in both physiological and psychological studies that habituation to stress occurs with repeated exposure to the stressor.(Flinn & England 1995; Okeeffe & Baum 1990)

Previous Diagnosis of a Relapsing-Remitting Condition

During the pilot study, the exclusion criterion was that no other medical condition was to have been diagnosed. However, it is known that having a diagnosis of epilepsy is associated with an increased likelihood of having another co-morbidity. (Gaitatzis et al. 2004; Wiebe et al. 2009) It therefore seemed unlikely, especially as the participants were being investigated for a potential cause of their symptoms, that a rigorous exclusion of all those who had another diagnosis would be possible.

In order to assess the stress experienced by the parent when their child was presenting with a potential diagnosis of epilepsy, the exclusion criterion was revised to mean a diagnosis of another condition that is relapsing and remitting in nature. This alteration to the meaning of the exclusion criterion was made as it was felt that a chronic co-morbidity is a stable condition and could be assumed to cause the same level of stress across the two questionnaire time points. The stress caused by another existing condition may cause a higher reported stress level at initial presentation, but this will then be standardised in the completed follow-up questionnaires. Therefore, the stress caused by the potential diagnosis of epilepsy can then be assessed. This is based on Spielberger's state-trait anxiety theory, where trait denotes the chronic condition that provides a similar level of anxiety. The potential epilepsy diagnosis would represent state anxiety, a transient period of increased anxiety that has been shown to be a function of stress.(Bedell & Roitzsch 1976)

A co-existing condition that was relapsing and remitting in nature represents a similar disease pattern to epilepsy.(Sillanpää & Schmidt 2006) Thus, it would not be possible to depict, by using the follow up questionnaires, as to which condition, the new onset epilepsy or the co-existing relapsing remitting condition, was causing the stress that was being experienced by the family.

4.3.7 Selection Procedure

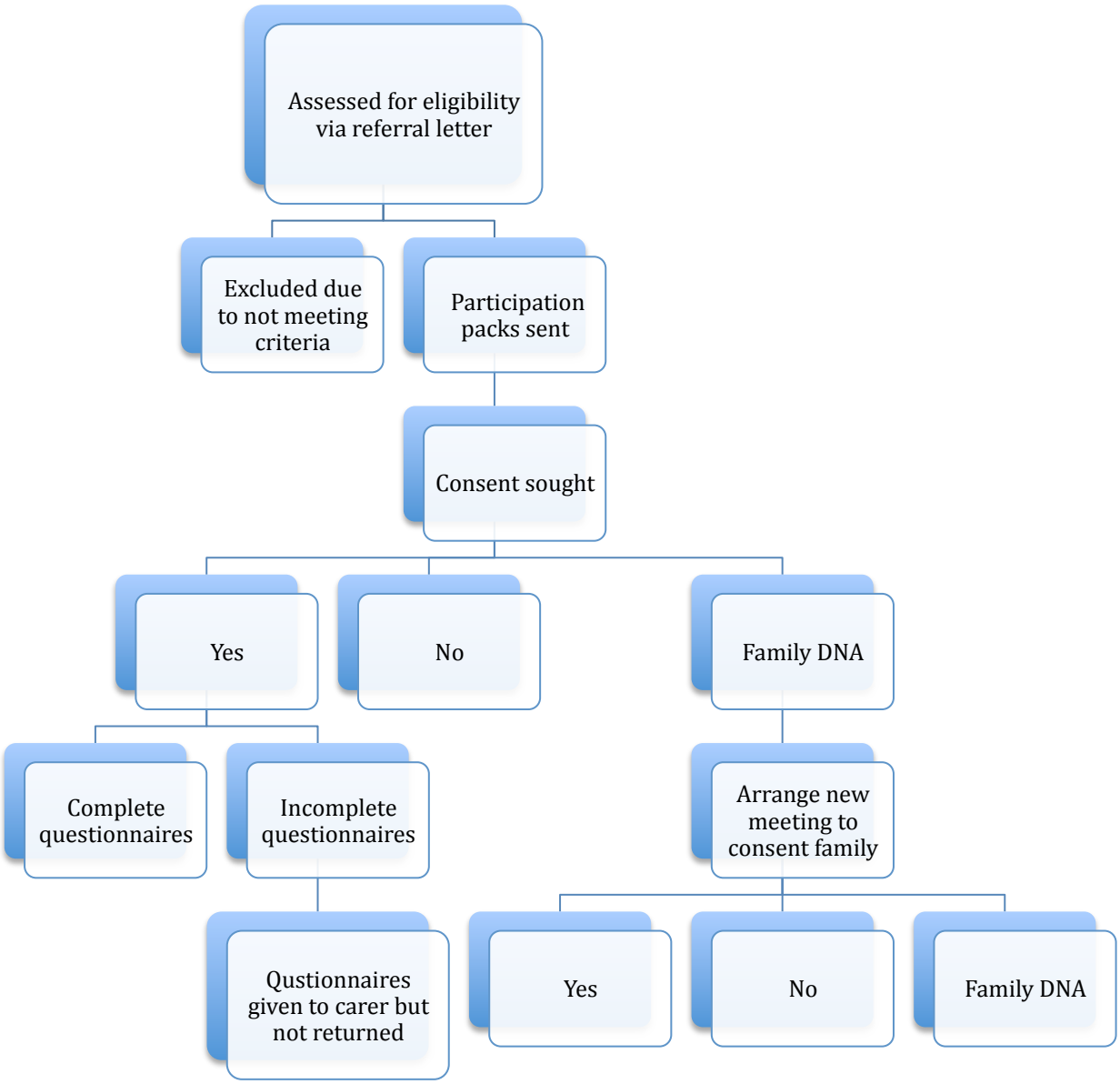
From September 2010 to June 2011, a postal participation pack was sent to families of children aged 0-16 years, who had been newly referred to Neurology or General Paediatric Outpatient Department at Alder Hey Hospital with a query diagnosis of epilepsy. The participation pack contained information for the parent and child, informing them of the study that was being conducted and asking if they would be interested in partaking in this research. An information sheet was sent to the child if they were over the age of 6 years. The information sheet given was age appropriate, modelled during the pilot study from examples supplied by the Research and Development Team at Alder Hey Trust and was further edited by the Local Ethics Committee to ensure that sufficient information was being provided. This produced two version of information sheets to be given according to the age of the child, both with the same content but with different wording: children aged 6-10 years received a version of the information sheet and those aged 10-16 years received another.

After being sent the participation pack, the family either replied to the participation pack and an appointment was made, or the family was met at their first clinic appointment. As the family had prior knowledge of the research and the information being sent was age-specific, informed consent could then be given to partake in the study.

The whole questionnaire pack was then completed at this time by the main carer and the child if aged 6 years or older.

4.4 Recruitment Process

Figure 8: Recruitment Process



Potential participants were approached for consent by meeting them at their outpatient clinic appointment. There were a few occasions where the main carer had responded to

the participation pack and therefore it was known if the family were or were not interested in taking part with the study. The outpatient clinic appointment was found out by using Meditech, the online database used at Alder Hey Hospital.

The family would then be approached whilst they were waiting for their clinic appointment, informing them of the study being performed and to discover whether or not they would be interested in taking part. Consent was sought from the main carer so that they and their child would be part of the study.

If interested, then they were informed of the length of time it would take to complete the questionnaires, that all information was made anonymous and that they would be approached to complete the final set of questionnaires 6 to 12 months later.

Following this information, the main carer and the child, if appropriate, signed the consent form and completed the questionnaire pack either before or after their clinic appointment in a separate room within the outpatient department.

There were a few occasions where the carer was not able to complete the questionnaires around that clinic appointment. When this occurred, one of two options was employed:

1. An alternative meeting was arranged to coincide with their next visit to Alder Hey Hospital. This was arranged only if a diagnosis of epilepsy had not been rejected nor confirmed and if the clinic appointment was within the next few weeks. This was to ensure that all participants still had a potential diagnosis of epilepsy. The participants were asked if they could arrive 30 minutes before their next scheduled clinic appointment so that the questionnaire pack could be completed. The setting for completing the questionnaire pack was in a separate room within the outpatient department.
2. The questionnaire pack was given to the carer and they returned all completed forms either with their next clinic appointment or via post to the Alder Hey Neurology Outpatient Department. When questionnaires were sent via the post, the questionnaires were explained to the carer before they left and contact details were given to them in case they had any questions regarding the questionnaires that needed clarifying. Consent was obtained from the carer to contact them a week after they were given the questionnaires to check that no problems had occurred in completing them.

From Figure 8 it can be seen that participation numbers were reduced in three ways:

- The family did not want to participate in the study and refused to take part when they were met at clinic.
- The family did not attend their outpatient clinic appointment.
- The family had agreed to take part in the study but questionnaires were not completed.

When a family did not attend their set outpatient clinic appointment, the strategy employed to try and recruit the patient was that they would be met again at their next outpatient appointment. This date was found through Meditech and the potential participants were met with the response being that they either agreed or refused to take part in the study. If the patient did not attend this new outpatient clinic date then it was felt that pursuing them further was not appropriate.

The second difficulty encountered when attempting to get completed questionnaire packs was that there were times that due to the carer's own time constraints, they were not able to complete all questionnaires at the clinic appointment.
When this occurred, a separate meeting was scheduled for their next visit to Alder Hey-the same conditions applied as mentioned previously (next clinic appointment within a few weeks time and diagnosis of epilepsy had not been confirmed nor refuted).
If there was not a visit to Alder Hey Children's Hospital that was scheduled imminently, the questionnaires were given to the carer to complete and then send back. The difficulties in both of these situations were that the family did not attend the next scheduled clinic appointment, or that the questionnaires were returned but only some of them were complete. If consent had not been gained to contact the carer and they had missed the clinic appointment, it was deemed inappropriate to contact them in an attempt to get the questionnaires complete.

4.5 Study Design

A cross-sectional study was carried between September 2010 and June 2011 at a single centre. All participants were asked to complete a battery of questionnaires at a single time point: when their child was presenting with a potential diagnosis of epilepsy. The outcome measures were pre-determined by a pilot study that was carried out during the academic year 2009/2010.

4.5.1 Choice of Study Design

With every research method, there are limitations and advantages. Previous studies that have examined stress in parents who have a child with a chronic disease, have consisted of study designs that used either a quantitative,(Putnick et al. 2008) qualitative (Mu 2008a) or a multiple research approach.(Cullen & Barlow 2004)

This study design is quantitative with questionnaires being given to parents or carers when they initially present with their child to an outpatient department with a queried diagnosis of epilepsy. The quantitative questionnaires gain an objective measure of carer stress, support and health; family functioning and future needs; carer and self-report of the child's quality of life. The benefits of a quantitative study design is that all findings can be generalised to the population and the effect of time upon carer stress can be compared as well as identifying the protective factors that help a family in bonadapting to their child being diagnosed with epilepsy.

As mentioned above, studies examining stress in parents have also used qualitative research methods. It is feasible to suggest that qualitative methods through interview or focus groups could have been used in this study as the data being presented within this thesis is from one time point. Focus groups would be able to provide an overview of carer's reasons for feeling stressed when presenting initially with their child. Interviews would provide a more detailed individual discussion of why the carer may feel stressed and highlight methods undertaken to help alleviate any burden they may feel. Therefore, both the quantitative and qualitative methods would be able to demonstrate associations between carer stress and other variables. It was decided not to undertake qualitative methods as causation of carer stress is trying to be achieved. As the data collected within this thesis is to be continued over a further two-year period in order to assess the effect of a carer's child diagnosis by comparing results before and after a diagnosis is given, questionnaires provide a standardised reliable measure. The

validity of qualitative research measures is dependent upon the researcher's expertise.(Leung & Savithiri 2009) As a different researcher is to be employed every year for data collection, interviewer bias may affect the quality of the collected information.(Appleton 1995)

Employing quantitative questionnaires will allow for a statistical comparison between the two time points of before and after a diagnosis is given to the child. It can then be assessed to see if a diagnosis of epilepsy affects the questionnaire scores more than the children who are not diagnosed with epilepsy.

By statistically assessing different factors that might contribute to family stress, the hope is that strong evidence will be collected that can be used to generate an intervention that will help reduce the experience of stress in families, optimising the family's wellbeing.

4.6 Variables and Outcome Measures Used

This is a multi-questionnaire study that is primarily completed by the child's main carer. If two carers are present, then the carer who looks after the child most during the week completes the questionnaire pack.

There are 9 questionnaires to be completed by the carer along with a demographic screening inventory.

Carers complete all 9 questionnaires if the child is 4 years or older.

If the child is under 4 years then the carer cannot complete the Strength and Difficulties Questionnaire as this has only been validated for children aged 4-16 years.(Goodman 2001)

There are two questionnaires that are completed by the child. However, this only occurs if the child is 6 years or older.

If the child is aged 6-16 years then they are able to complete the Pediatric Quality of Life Inventory (PedsQL 4.0).(Varni, Seid & Kurtin 2001)

If the child is aged 7-16 years, then they are able to complete the Self Image Profile: 7-11 years complete the children version (SIP-C), 12-16 years complete the adolescent version (SIP-A).

These age criteria were created due to the questionnaires only being validated for these ages.(Butler 2001)

All the outcome measures are generic and non are specific to epilepsy. The reason for this is the diagnosis of epilepsy is only potential and some will be later told that they do not have epilepsy. It was therefore deemed inappropriate for carers or children to complete epilepsy-specific questionnaires and generic ones were given instead.

A summary of the questionnaires completed by the carer and child can be seen in Table 9. Table 10 tabulates the questionnaires used and the variables that it is serving to measure.

Table 9: List of questionnaires completed by the carer and the child.

Carer			Child 6 years	Child 7-11 years	Child 12-16 years
Pediatric Inventory for parents (PIP)	Internal-External locus of control	Strengths and Difficulties Questionnaire (SDQ)	Pediatric Quality of Life Inventory (PedsQL 4.0)	Self Image Profile for Children (SIP-C)	Self Image Profile for Adolescents (SIP-A)
Semi-structured stress questionnaire	Family Adaptability and Cohesion Evaluation Scales, version IV (FACES-IV)	Brief COPE Inventory		Pediatric Quality of Life Inventory (PedsQL 4.0)	Pediatric Quality of Life Inventory (PedsQL 4.0)
General Health Questionnaire 28 (GHQ-28)	Family Needs Survey (FNS)	Family support scale (FSS)			

4.6.1 Alternative Variable Measurement Tools that Could have been Used

A review was undertaken of the commonly used measures in studies examining parental stress when their child is diagnosed with epilepsy. This can be seen in Appendix 2. The review documented the study design, outcome measures used as well as the study's findings. This was undertaken out of interest so to understand how this present study's methodology compared with those that had a similar aim. This study's methodology had already been decided upon when this years work was undertaken, however the review in Appendix 2 shows the variety in alternative self-reported questionnaires that could have been used. Upon review, similar reliability and validity scores have been reported for the questionnaires used in this study compared to alternatives. The alternative measures that could have been used display that there is a wide range in the number of items within each questionnaire. This implies that there is

a range in the length of time that it would take to complete each questionnaire. The questionnaires used within this methodology were found from the pilot study to all took a maximum of 5 minutes to complete each, making the time taken to complete the battery of questionnaires to be between 30-40 minutes.

The Pediatric Inventory for Parents (PIP) reported the strongest reliability scores compared to alternative measures of stress, strengthening the use of this questionnaire. This can be seen in Appendix 2. This is important to note, as the PIP will be used in order to assess associations between variables and carer stress. The methodology that was used will additionally be considered within the main discussion.

Table 10: Variables Being Assessed and the Questionnaires Used

Variable		Concept	Instrument	Normal values
Carer stress		Understand whether daily illness related stresses have been occurring to carers in the past 2 weeks	Pediatric Inventory for Parents (PIP)	No clinically determined point that indicates stress. Using total score of 90 as cut off point, based on previous study findings.(Guilfoyle et al.)
		Understand reasons behind the impact that a potential diagnosis of epilepsy has on carer stress	Semi-structured Stress Questionnaire	Not clinically validated Using total score of 10 as cut off point. (Based on conversation with Dr Andrew Curran)
Carer Coping • Problem-solving strategies • Emotional strategies • Dysfunctional strategies		Assess the different coping strategies employed by the carer in relation to the potential diagnosis of epilepsy. 28 items to create 3 subgroups: dysfunctional 12-item; emotional 10-item; problem focused 6-item.	Brief Coping Orientations to Problems Experienced (COPE) Scale.	Means and standard deviations(Cooper, Katona & Livingston 2008) Dysfunctional 16.1 (4.4); Emotional 19.4 (5.3); Problem-focused 11.7 (4.5).
Carer Health		Assess whether the potential diagnosis of epilepsy in their child affects carer health	General Health Questionnaire-28 (GHQ-28)	Cut-off value of 5 to indicate poor health(Goldberg et al. 1997)
Carer locus of control		Examine whether there is a difference in carers conception of control; do they believe they have control over their life events (internal) or that they occur due to fate (external)	Internal-External Locus of Control	Median value taken to indicate the two groups.(Parkes 1984) Our median value was 12: 0-12 = Internal 13-23 = External
Carer Resources	Carer Support • Formal kinship • Informal kinship • Social groups • Professionals • Professional groups	Examine the different sources of support and the perceived helpfulness of the support that has been viewed as being available to the carer. Total score is generated from the 5 subscales to indicate overall perception of support available to family	Family Support Scale (FSS)	No normal values reported by author. Higher scores indicate more support perceived.(Hanley et al. 1998) Total support score mean and standard deviation of 27.25 (11.2) was reported from Taylor et al.(Taylor & Others 1993)
	Carer Needs • Resource • Information • Counselling	Understand which needs are viewed by the carer as being a 'definite need' now that their child has a potential diagnosis of epilepsy.	Family Needs Survey (FNS)	Higher scores indicate higher unmet needs. (Trute & Hiebert-Murphy 2005)

Variable	Concept	Instrument	Normal values
Child's behaviour -proxy report • Emotional problems • Conduct problems • Hyperactivity • Peer relationship problems • Prosocial behaviour • Impact	A behaviour screen of the carer's child. A total score is generated using the 5 subscale scores. Additionally, an impact score is included to indicate the perceived impact on home, recreation and education as a result of their child's behavioural difficulties.	Strengths and Difficulties Questionnaire (SDQ)	Normal total mean and standard deviation score for parent report is 8.4 (5.8).(Goodman et al. 2000)
Family Functioning • Family Communication Scale • Family Satisfaction Scale (FSS)	Understand the different levels of family togetherness and ability to respond to change. Assess the quality of communication between family members Assess the satisfaction of the family	Family Adaptability and Cohesion Evaluation Scale –IV (FACES-IV)	Total circumplex ratio describes a summary of mean balanced to mean unbalanced score. Higher the ratio is above 1, the more healthy the family is. Most between 0-2.(Olson, Gorall & Tiesel 2006) Communication mean and standard deviation 36.2 (9.0). FSS mean and standard deviation 37.5 (8.5).(Olson, Gorall & Tiesel 2006)
Child's self image and self esteem	To assess whether the children with the potential diagnosis of epilepsy have a positive (SI +ve) or negative image (SI –ve) and what their self esteem (SE) is like.	Self Image Profile for Children (SIP-C) Self Image Profile for Adolescents (SIP-A)	User guide reports mean scores depending on age and gender.(Butler 2001) Mean and standard deviation of scores for SIP-C and SIP-A. (Bellew, Haworth & Kay 2011) SIP-C: SI +ve 50.67 (8.78) SI –ve 27.44 (8.96 SE 33.0 (16.03) SIP-A: SI +ve 40.60 (6.88) SI –ve 34.40 (11.76) SE 32.80 (14.82)
Child's quality of life	To assess the child's perception of their quality of life within the past month	Pediatric Quality of Life Inventory 4.0 (PedsQL 4.0)	Mean and standard deviation of normal total score from child report was reported to be 83.0 (14.79)(Varni, Seid & Kurtin 2001) and 83.84 (12.65) from a population of 5480.(Varni, Limbers & Burwinkle 2007)
Carer demographic screen	Overview of carer's family structure, their level of education, weekly finance and number of dependencies.	Demographic Information Sheet	Not applicable
Child's fit history	A brief overview of the episodes being experienced: fit frequency, if occurred in a public place, thoughts on condition.	Demographic Information Sheet	Not applicable

Figure 9: Schematic Diagram of the Studied Variables and the Measurement Tools Used.

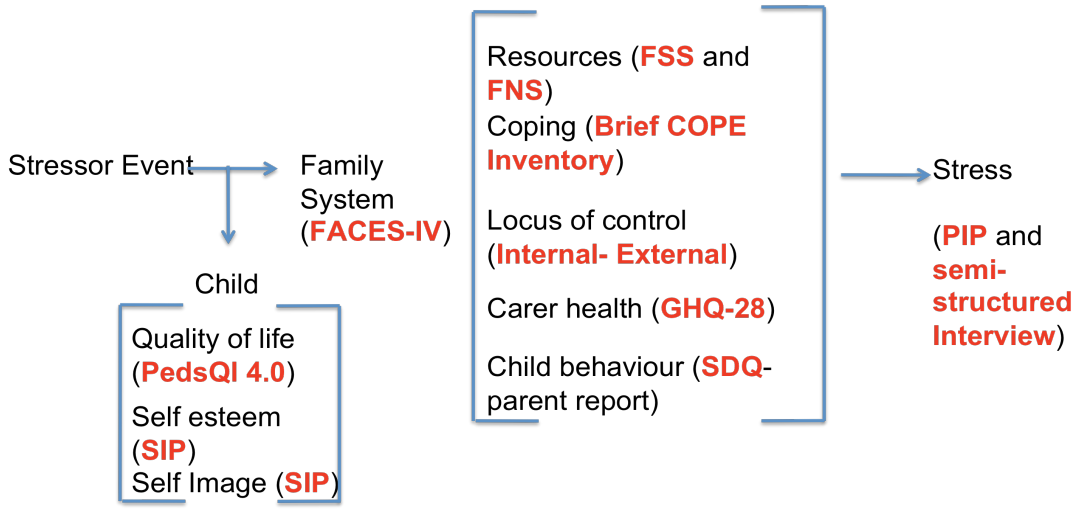


Figure 9 describes the rational behind each of the questionnaire used. This figure is based on the models generated within the family stress theories. This study was not based on a specific family stress theory but has used the conceptual idea that certain variables may act as a mediator to stress.

From the family stress theories, family capabilities incorporates coping, family functioning and family resources and these have been posited as variables that have the ability in allowing the family to adjust to a stressor or crisis.

From the literature review and critical appraisal, it was clear that certain variables have been linked to affecting carer stress. These are the carer's own health, their child's behaviour, the coping strategies used, the family functioning and the resources that were available to them. Therefore, the methodology was developed to assess each of these variables as well as the carer stress. Using this schematic diagram of the methodology as depicted in Figure 9, stress is the independent variable, with all other variables that are completed by the carer being the dependent variables.

It has also been shown within the literature review that being diagnosed with a chronic condition has the ability to affect the child's quality of life and their self-esteem. Therefore, these two variables were additionally incorporated into the study design although they will not be contributing to the assessment as to whether or not they affect carer stress.

4.7 Variable Measurement Tools

4.7.1 Carer Stress

4.7.1.1 Pediatric Inventory for Parents (PIP) (Streisand et al. 2001)

Carer stress was assessed using the Pediatric Inventory for Parents (PIP). This is an illness specific measure of parental stress, developed in 2001 using reported stress levels from 126 parents of children with cancer. It was intended to be a tool that could be applied to different illness groups due to the general nature of the illness-specific items.

The PIP is a 42-item inventory that asks parents to rate, using a Likert scale of 1-5 (1= 'Not at all,' 5 = 'Extremely'), how frequently the events have occurred within the past 14 days and how difficult it has been for them. This formulates two total scores for the frequency of the items (PIP-f) and a score for the difficulty associated with the events (PIP-d).

Within each score, four domains have been identified so to highlight what aspect carers find most stressful. The domains are: communication (9 items), emotional distress (15 items), medical care (8 items), and role function (10 items).

Reliability of the PIP has only been reported using Cronbach's coefficient alpha, demonstrated high internal consistency (PIP-f, $\alpha = 0.95$; PIP-d, $\alpha = 0.96$) (Streisand et al. 2001) and this has since been confirmed in a recent study examining stress in parents of children with inflammatory bowel disease (PIP-f, $\alpha = 0.96$; PIP-d, $\alpha = 0.95$).(Guilfoyle et al.)

Validity of the PIP was established by comparing the total frequency and difficulty PIP scores to existing validated questionnaires: State-Trait Anxiety Inventory for Adults (information about the questionnaire taken from (Barnes, Harp & Jung 2002)) and the Parenting Stress Inventory-Short Form (PSI-SF). (Abidin & Wilfong 1989)

Streisand et al found significant correlations between PIP-f and PIP-d and state anxiety scores (rs = 0.62 and 0.60 respectively) and slightly lower but significant correlations with the parent subscale of the PSI-SF (0.38). Both results had a significance level of $p<0.01$.(Streisand et al. 2001) Ohleyer et al also demonstrated correlations between the total scores for the PIP-f and PIP-d against the total score for PSI-SF. This was shown using Pearson product-moment correlation; $r=0.52$ and $r=0.49$ respectively. The Child Behavioural Checklist (CBCL) was also correlated against PIP-f and PIP-d, showing

similar significantly positive correlations for the internalising (0.47, 0.46) and externalising (0.53, 0.48) sub-domains of the CBCL ($p<0.01$).(Ohleyer et al. 2007)

A clinical cut-off score of the PIP to indicate whether stress being experienced by the carer is significantly high compared to a normal population, has not yet been developed.(Guilfoyle et al.) Despite this limitation of the questionnaire, it has been used in various other studies to examine carer stress when a child has been diagnosed with different conditions. This can be seen in Table 11.

The mean scores obtained from the 7 different studies, gave an average PIP-f of 92.4 and PIP-d of 88.5. Therefore, a score >90 was felt to be justified as a means of signifying whether or not the carer was experiencing significant stress. As there are two domains of the stress score, it was felt that the overall stress score for each carer should be represented by an average of both their PIP-d and the PIP-f score. If this was >90, then the carer was categorised as being stressed. As the average PIP-d and PIP-f scores from the 7 previous studies produced a combined score of 90.45, it was felt that an average carer stress score using their total PIP-d and PIP-f scores, could be used as a means of categorising whether or not the carer is overall stressed. This therefore provides an overview of how many carers are stressed when initially presenting with their child. The advantage of this is that it uses the whole overview of carer stress; accounting for stress caused by both the frequency and perceived difficulty of the health-related events. The disadvantage is that it might not accurately estimate the numbers of carers who are stressed by either over or under estimating those classified as being stressed when only one domain states that they are stressed whilst the other is under the cut-off value of 90. It shall be analysed to assess the number of carers who have been potentially misclassified due to the method of averaging the results from both domains.

Table 11 highlights that the internal consistency of the PIP has been demonstrated to be very high in the studies that have used the measure and also documents the study's findings that were then used to create the categorisation that shall be used in this study to indicate whether significant stress was being felt by the carers.

Table 11: Studies that have used the PIP and their findings

Study	Condition	Sample size	Results Mean (standard deviation)		Reliability- Internal consistency
			Total PIP-f	Total PIP-d	
(Streisand et al. 2001)	Cancer	126	94.0 (33.3)	112.4 (35.1)	PIP-f, α =0.95 PIP-d, α =0.96
(Logan, Radcliffe & Smith-Whitley 2002)	Sickle cell disease	70	105.4 (27.3)	91.1 (33.0)	Not reported
(Streisand et al. 2005)	Diabetes	134	89.3 (26.0)	78.1 (26.1)	PIP-f, α =0.94 PIP-d, α =0.95
(Ohleyer et al. 2007)	Obesity	72	98.0 (34.4)	91.9 (34.2)	PIP-f, α =0.96 PIP-d, α =0.96
(Mednick et al. 2009)	Bladder Exstrophy	20	89.8 (23.2)	90.5 (28.9)	PIP-f, α =0.93 PIP-d, α =0.94
(Wagner et al. 2010)	Epilepsy (pre-intervention)	9	86.1 (24.6)	77.6 (21.7)	Not reported
(Guilfoyle et al.)	Inflammatory Bowel Disease	62	84.4 (27.9)	78.2 (25.2)	PIP-f, α =0.96 PIP-d, α =0.95

4.7.1.2 Semi-Structured Stress Questionnaire

The semi-structured stress questionnaire was created by Andrew Curran, which is based on the 'Family Burden of Injury Interview' (FBII) created in 1995 by Taylor et al.(Burgess et al. 1999)

The FBII is a 31 item structured interview, designed from 99 parents of children who had suffered a traumatic brain injury.(Burgess et al. 1999) The aim of the FBII is to assess injury-related stress as perceived by the parent. This includes items such as their concern about the child and their partner's reaction to the condition.(Taylor et al. 1999) These items are both included within the semi-structured stress questionnaire used within this study.

Each item within the FBII is to determine whether the parent is experiencing burden, and if it is a cause of concern to the parent, they rate the level of stress on a Likert scale of 0-4; 0 indicating no stress, 4 indicating an extremely stressful issue. For each item that is identified as an area of concern to the parent, they are then able to describe the issue in more detail, which is recorded by the interviewer.

Following a conversation between Andrew Curran and Jerry Taylor, the creator of the FBII, a FBII score of >10 was advised as signifying more than normal levels of stress. (Information gathered from personal communication with Andrew Curran)

The semi-structured stress questionnaire has not been validated nor checked for reliability, but it has been used previously piloted in a group of 64 children with disabilities, used within a therapeutic environment. (Unpublished data: Andrew Curran) The FBII was assessed as an indication for the semi-structured stress questionnaire's reliability and validity. This can be seen in Table 12.

Table 12: Reliability and Validity of the FBII (Burgess et al. 1999)

Reliability of the FBII total score	
Cronbach's alpha	0.90
Split-half reliability	0.80
Test-retest	r=0.64 (baseline to 6 months) r=0.52 (baseline and 12 months)
Validity of the FBII	
BSI (maternal psychological distress)	r=0.36, p<0.01 at baseline
IOF-G (impact of child's health status on family)	r=0.49, p<0.01 at baseline
FAD-GF (general family function)	r=0.38, p<0.01 at 6 month follow up

The semi-structured stress questionnaire is structured in the same way as the FBII. There are 25 items that the carer responds to as to whether it is considered a source of stress. If it is, the carer rates on a Likert scale of 0-4, the amount of stress caused by each of the 25 items. This gives a range of 0-100 for the total stress score, with a total score of >10 said to signify increased carer stress compared to a healthy population.

With each question that is rated, an additional qualitative element is included as a chance for the carer to further explain why or why not them item has caused stress to them. The qualitative data will not be presented within this thesis.

Although the semi-structured stress questionnaire has not been validated whilst the FBII has been, it was decided that the semi-structured stress questionnaire was a more appropriate measure to use due to the phrasing of the questions. The reason for this was that the FBII was formulated specifically for traumatic head injury with the questions inquiring after stress caused by the injury, whilst the semi-structured stress questionnaire is specific to stress caused by the seizures and the query diagnosis of epilepsy. The semi-structured stress questionnaire has a similar structure to the FBII, with similar questions and the same scoring system. This makes it feasible to extrapolate the validation data from the FBII to the semi-structured stress questionnaire. However, it may be considered inappropriate to extrapolate the validation data from the FBII to the semi-structured stress questionnaire, as it is a different questionnaire, aimed at another condition that may produce a different stress response to that seen in epilepsy.

4.7.2 Carer Health

4.7.2.1 General Health Questionnaire-28 (GHQ-28) (Goldberg & Hillier 1979)

The General Health Questionnaire (GHQ) is a self-administered screening questionnaire that was designed to detect a psychiatric disorder within the general population, (Goldberg & Hillier 1979) and has been used within the community and non-psychiatric settings such as general medical outpatient departments.(Vallejo et al. 2007)

The GHQ-28 is a 28-item questionnaire that was created from the original larger version of the GHQ containing 60 items.(Goldberg & Hillier 1979) From the 28 items, four different subgroups can be created, each consisting of 7 items: somatic symptoms, anxiety and insomnia, social dysfunction and depression.

It assesses the psychological state of the respondent within the last few weeks compared to their usual state and is therefore a good tool to detect short-term change.(Vallejo et al. 2007)

It is additionally the only version of the GHQ that contains a specific anxiety subgroup.(Meades & Ayers) Due to these factors and as the questionnaire has been shown to have good validation data within the general population,(Friedrich et al. 2011) it was deemed an appropriate instrument to screen carers for disorders such as anxiety and depression, especially as it has been shown in previous studies that there is an association between carer mental health and children with chronic conditions.(Boman, Lindahl & Björk 2003; Chiou & Hsieh 2008b)

There have been four different methods suggested to score the GHQ: Bimodal (0-0-1-1), Likert (0-1-2-3), Modified Likert (0-0-1-2) and the Chronic scoring method (0-0-1-1 for positive worded items and 0-1-1-1 for negatively worded items).(Friedrich et al. 2011)

Goldberg suggested that the Bimodal format was best suited for the GHQ-28 (Goldberg & Hillier 1979; Goldberg & Williams 1988) and in a recent review of the different scoring methods available, Friedrich found that the Bimodal scoring method was the most accurate for screening non-psychiatric patients.(Friedrich et al. 2011) As Goldberg recommends the Bimodal scoring for identifying cases,(Goldberg &

Williams 1988) this method will be used, as it additionally will increase the scope of future work possible by using the database.
Using the Bimodal scoring method, a total GHQ score of 0-28 is possible, with a higher score indicating ill health.

Originally a cut-off of 4/5 was recommended,(Goldberg & Hillier 1979; Goldberg & Williams 1988) but following from Goldberg's 1997 study, a cut-off value of 5/6 is recommended to identify cases.(Goldberg et al. 1997)
This shall be the score used to determine whether or not this study's population is reporting ill health. The validity and reliability values are reflecting the cut off score that shall be used, 5/6.
Reliability has been displayed using Cronbach's alpha coefficient, and has been shown in a study involving 80 Polish subjects who were approached in a primary care setting. They found the internal consistency to be 0.911.(Makowska et al. 2002) A similar finding was demonstrated in a student population of 100 people by demonstrating an internal consistency of 0.9.(Vallejo et al. 2007) Vallejo et al additionally assessed the test-retest reliability and found that after 17 days, total score correlation was 0.69 ($p<0.001$). (Vallejo et al. 2007)

The validity of the GHQ-28 has been demonstrated by a large international study conducted by Goldberg et al. A population of 5438 was recruited from general health practices that were diagnosed with a variety of psychological disorders. Comparing the results generated by the GHQ-28 to that by the Composite International Diagnostic Instrument (CIDI), they found that a cut-of value of 5/6 resulted in an overall 79.7% sensitivity rate and 79.2% specificity rate. The positive predictive value using the GHQ-28 was 54.7.(Goldberg et al. 1997)
Makowsak et al found that when comparing the GHQ-28 threshold score of 5/6 to the diagnosis as determined by the CIDI in a sample size of 80, sensitivity was 59% and specificity was 75%. Overall misclassification rate was 30%.(Makowska et al. 2002)

4.7.3 Carer Coping

4.7.3.1 Brief Coping Orientations to Problems Experienced (COPE) Inventory (Carver 1997)

The Brief COPE Inventory is a 28-item self-report questionnaire. It was formulated from a longer version of the COPE Inventory that contained 60 items. The Brief COPE consists of 14 scales, with each scale consisting of two items. The scales are: active coping, planning, positive reframing, acceptance, humour, religion, emotional support, instrumental support, self-distraction, denial, venting, substance use, behavioural disengagement and self-blame.(Carver 1997)

Each of the 28 items represents a type of coping strategy. The coping strategies being assessed are a mixture of those that seem potentially dysfunctional as well as adaptive responses.(Carver 1997) For each item, a rating scale of 1-4 is employed as a means of assessing the degree to which the coping strategy has been used since it was mentioned that their child might have epilepsy.(Segal, Hook & Coolidge 2001)

The responses possible are:

1- I haven't been doing this at all
2- I have been doing this a bit
3- I have been doing this a medium amount
4- I have been doing this a lot.

The responses were meant to be within a format that was situational and retrospective, as suggested by Carver,(Carver 1997) to accurately assess coping strategies specific to the stressor; the potential diagnosis of epilepsy. The Brief COPE is an effective coping inventory as it assessed many different coping strategies as well as the situational coping employed by parents within this specific period of time.(Hastings et al. 2005)

The Brief COPE Inventory has additionally been assessed for its reliability and validity. Carver stated that the internal consistency for each subscale was >0.50.(Carver 1997)

Amoyal et al used the Brief COPE to a group of 362 participants who had sustained a burn injury. They found that the internal consistency varied between 0.557-0.855 when grouped into 7 types of coping styles. Using the same 7 groups, test-retest after 6 months demonstrated a reliability of 0.164-0.626.(Amoyal et al. 2011) Hastings et al found from their study that involved 135 parents of children diagnosed with autism,

the internal consistency of the Brief COPE to vary between 0.68-0.82 when the Brief COPE was used to form four groups.(Hastings et al. 2005) Amoyal et al demonstrated the construct validity of the Brief COPE, by finding an association with the short form health index. They found that a negative correlation occurred between avoidance coping and the mental health subscale of the short form health index. Acceptance, part of the emotional focused coping strategies was found to be positively associated with less distress at discharge and 12 months afterwards.(Amoyal et al. 2011)

As shown by the studies mentioned already, Carver intended the Brief COPE Inventory to be flexible in its application and the grouping of the coping strategies.(Carver 1997) Carver originally described within the full COPE Inventory that there are three main groups of coping strategies, being problem-focused, emotion-focused and dysfunctional.(Carver, Scheier & Weintraub 1989)

A study by Cooper et al grouped the Brief COPE Inventory into three different ways of coping. This present study groups the Brief COPE items in the same way:(Cooper, Katona & Livingston 2008)

Emotional-focused strategies: Acceptance, Emotional support, Humour, Positive Reframing, Religion

Problem-focused strategies: Active coping, Instrumental support, planning

Dysfunctional strategies: Behavioural disengagement, Denial, Self-distraction, Self-blame, Substance use, Venting.

The scores for each group are: emotional-focused (10-40), problem focused (6-24) and dysfunctional (12-48). Cooper et al demonstrated good internal reliability for these groups from a sample of 125 caregivers. The Cronbach alpha coefficients were 0.72, 0.84 and 0.75 for emotional, problem-focused and dysfunctional coping strategies respectively.(Cooper, Katona & Livingston 2008) Test-retest reliability also demonstrated good stability over a one and two year period. Baseline to one year correlation was $r=0.67$ and baseline to two year was $r=0.54$; $p<0.001$. (Cooper, Katona & Livingston 2008) Cooper et al additionally demonstrated the construct validity of these three groups, finding that emotional focused coping strategies correlated significantly with the amount of social support, dysfunctional coping was correlated with higher avoidant attachment scores and problem focused with activities of daily living functioning.(Cooper, Katona & Livingston 2008)

4.7.4 Carer Locus of Control

4.7.4.1. Internal-External Locus of Control(Rotter 1966)

Rotter created the Internal-External Locus of Control questionnaire in 1966; a self-report questionnaire designed to categorise an individual's locus of control as either internal or external.(Rotter 1966)

The questionnaire consists of 29 items. 23 of the items contribute to the overall locus of control score, with 6 items that do not. The 6 items that do not contribute to the overall score are called 'filler items' and were placed into the questionnaire in order to maintain ambiguity. (Beretvas et al. 2008)

Each item within the questionnaire has two statements, either statement 'a' or statement 'b'. Each statement measures the individual's belief about the nature of the world.(Rotter 1966) The subject who is completing the questionnaire must indicate as to which of the two options they agree with more. This is what Rotter describes as a forced-choice format.(Rotter 1990)

The scoring of the questionnaire is based upon the response given for each statement. The 23 items that contribute to the overall score are then scored either 0 or 1. A score of 1 is given to the statements that indicate an external locus of control.(Rotter 1975) Therefore, the higher the overall score, the more external that individual's locus of control is. Conversely, the lower the score, the more internal that individual's locus of control is.

Determining where the cut-off is in order to distinguish the two categories is not set; this is because Rotter describes the locus of control as being situation specific and should be viewed as a continuum with the lowest and highest scores indicating where the individual's locus of control lies.(Rotter 1975) However, studies that have used Rotter's Internal-External Locus of Control questionnaire have used the median value of the total sample as a means of distinguishing the two groups.(Beretvas et al. 2008; Parkes 1984; Renn & Vandenberg 1991) This shall be the technique employed within this study.

Rotter stated that the internal consistency for the scale was 0.69-0.73.(Rotter 1966) A systematic review of 72 studies, of varying sample sizes (n=14-7439), found the average internal consistency being an alpha coefficient of 0.663. Within the same review, 14 studies reported test-retest reliability estimates, and these ranged from 0.53-0.86 with the average length of time being 4 weeks.(Beretvas et al. 2008)There have

been attempts at determining the convergent validity of the Internal-External scale, displaying a correlation between Reid and Ware's fatalism subscale (0.55) and Levenson's Chance subscale (0.45).(Goodman & Waters 1987)

4.7.5 Family Functioning

4.7.5.1 Family Adaptability and Cohesion Evaluation Scale IV (FACES IV)(Olson 2011)

The Family Adaptability and Cohesion Evaluation Scale (FACES) is a 62 item, scored on a Likert scale of 1-5, self-report assessment questionnaire that assesses family functioning, based on the Circumplex Model.(Franklin, Streeter & Springer 2001; Olson 2011) The Circumplex Model was created in an attempt to map the different ways that a family functions, achieved by incorporating three concepts: family cohesion, flexibility and communication.(Olson 2011; Olson 2000)

Olson defines each concept as:(Olson 2011; Olson 2000; Olson & Gorall 2003)

- Cohesion is the emotional bond between family members.
- Flexibility is the quality and expression of leadership, role assignation and organisation.
- Communication is the positive communication that exists between family members.

For a family to have a 'healthy' level of function, a balance between cohesion and flexibility is needed.(Olson 2011; Olson 2000) It has been shown that in times of stress, families alter the way that they function by altering either their family's cohesion or flexibility.(Olson & Gorall 2003) This premise is based on families being viewed as organised systems that through effective communication and role assignment, strive to maintain balance.(Alderfer et al. 2008) Therefore, in a time of stress, families may begin to function more poorly, which is displayed by either being unorganised with ineffective communication between family members, or by becoming very regimented with inadequate communication.(Alderfer et al. 2008)

Thus, by including the FACES IV within the study, it is providing a general measure of family functioning,(Alderfer et al. 2008) which allows an assessment for the impact of a potential diagnosis of epilepsy is having upon family function.

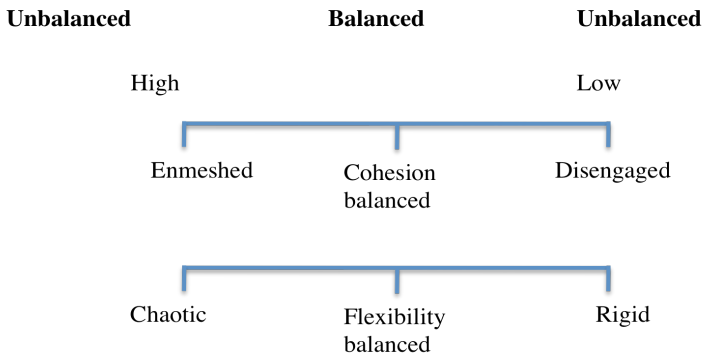
FACES IV was developed from the three previous FACES instruments that have existed, with the present version of FACES, FACES IV, occurring so that an accurate assessment of extremes of the scales cohesion and flexibility could occur as adjustment

was found to be linearly related to these extremes.(Franklin, Streeter & Springer 2001; Olson 2011)

The end result has been that FACES IV has six dimension; two scores indicating a state of balanced family function, and four scores indicating a state of unbalanced family function.(Olson 2011) This can be seen in Figure 10.

The six separate dimensions of the family's cohesion and flexibility are generated from the raw scores of 42 of the total 62 items within FACES IV: 7 items assess each dimension.

Figure 10: The Six Outcomes Assessed for Family Cohesion and Flexibility(Olson 2011)



As an overview of the family functioning is wanted for this study, the Circumplex Total Ratio will be used as a means of comparing the family's level of functioning. The Circumplex Total Ratio is a measure of the average balanced scores (balanced cohesion and flexibility) divided by the average unbalanced scores (enmeshed, chaotic, disengaged and rigid), and it is this score that generates how healthy the family function is.(Olson, Gorall & Tiesel 2006)

A Circumplex Total Ratio less than 1 signifies an unbalanced system with family functioning not at its optimum, a ratio of 1 indicates equal level of balance and unbalance in the family system and a ratio above 1 describes a more balanced and healthier family system.(Olson 2011)

The remaining 20 items within the FACES IV assesses the health of the family communication via the Family Communication Scale (FCS) and the family's satisfaction via the Family Satisfaction Scale (FSS).(Olson & Gorall 2003), (Alderfer et al. 2008) Both scales are assessed by 10 items each, with each item rated on a Likert scoring of 1-5; 1 = strongly disagree and 5= strongly agree. These scores, ranging from

10-50 for each scale, can then be converted into percentages that further correspond to 5 categories, ranging from 'Very Low' to 'Very High'.

The reliability of FACES-IV has been demonstrated through assessing the internal consistency using Cronbach's alpha and test-retest correlation using Pearson product-moment correlation. The validity of the FACES-IV has been demonstrated by assessing the predictive power and the convergent validity by comparing scores against the Family Assessment Device (FAD), the Self-report Family Inventory (SFI), the family support subscale on the Social Support Behaviour scale (SSB) and the family relationship problems subscale on the Multi-Problem Screening Inventory (MPSI). The results are tabulated below.

Table 13: Reliability and Validity from Studies Assessing FACES-IV

Study	Sample size and population	**Reliability**		**Validity**	
		Internal consistency (a)	Test-Retest	Concurrent/predictive	Convergent
(Olson 2011)	556 students and families	**Disengaged**: 0.87 **Enmeshed**: 0.77 **Rigid**: 0.82 **Chaotic**: 0.86 **Cohesion**: 0.89 **Flexibility**: 0.84 **FCS**: .90 to .94. **FSS**: .90 to .94.	**FACES-IV** over 3 weeks *r*s = 0.83 - 0.93 **FCS** (time not specified) *rs* = 0.86 **FSS** (time not specified) *rs* = 0.85	Is able to determine families that are "healthy" and "unhealthy". Families falling in the unbalanced groups show poorer family quality and greater stress. Predictive accuracy of 92% when comparing clinical to non-clinical groups	**Disengaged**: r = -0.81 with SFI. r= -0.27 to -0.82 with FAD. **Cohesion**: r = 0.88 with SFI. r= 0.40 to 0.85 with FAD. **Flexibility**: r =0.68 with SFI. r= 0.21 to 0.63 with FAD. **Chaotic**: r = -0.62 with SFI. r= -0.20 to -.63 with FAD. **Satisfaction**: r= 0.86 with SFI. r= 0.84 with FAD.
(Franklin, Streeter & Springer 2001)	105 expectant mothers	Enmeshed = 0.75 Disengaged = 0.79 Rigid = 0.65 Chaotic = 0.76	Not reported	Predicting accuracy: Disengaged- 93.2% Chaotic- 86.4% Rigid- 69% Enmeshed- 68% Total circumplex score- 84.1%	Against total FACES IV score: FAD = -0.362 MPSI = 0.503 SSB = -0.301
(Marsac & Alderfer 2010)	162 families with child diagnosed with cancer	Cohesion = 0.78 Flexibility = 0.74 Disengaged = 0.74 Enmeshed = 0.65 Rigid = 0.7 Chaotic = 0.72	Not reported		Using mother's scores compared against FAD. Cohesion: r=0.59 Flexibility: r=0.26 Disengaged:r=-0.47 Emneshed: r=-0.09 Rigid: r=-0.06 Chaotic: r=-0.54

4.7.6 Family Resources

4.7.6.1 Family Support Scale (FSS)(Dunst, Trivette & Cross 1986)

The Family Support Scale (FSS) is an 18-item inventory designed to measure the amount of perceived support that is provided to the carer.(Dunst, Trivette & Cross 1986) It uses a five point scale from 1 'not at all helpful' to 5 'extremely helpful'. There is an additional option of 'not applicable' scoring 0 on the scale for support that is not available to the family.(Taylor & Others 1993) Summing the scores from the 18 items provides a total score, ranging from 0-90, depicting the number of sources of support available to the carer within the past 3-6 months.(Dunst, Trivette & Cross 1986; Hanley et al. 1998)The higher the score, the more support the family receives.(Glenn et al. 2009; Taylor & Others 1993)

Using the 18-item FSS, different researchers have created different subgroups as a means of mapping the helpfulness of the support that is being given to the family.(Hanley et al. 1998; Hastings et al. 2002) Dunst originally created six subgroups of support but altered it to five subgroups.(Dunst, Trivette & Cross 1986) Taylor altered the five subgroups to only four different groups of support.(Taylor & Others 1993)However, Hanley et al found that a five factor made more clinical sense compared to other suggested subgroups.(Hanley et al. 1998) This present study is using five subgroups as formulated by Dunst (Dunst, Trivette & Cross 1986) and which has been used in other studies, range of scores shown in the brackets:(Hanley et al. 1998; Hassall, Rose & McDonald 2005) formal kinship (0-15), informal kinship (0-25), social groups (0-10), professionals (0-20) and professional groups (0-20).

Hanley et al demonstrated good internal consistency, 0.85 and test rest correlation of 0.73 for the total FSS score using a population size of 244.(Hanley et al. 1998) Dunst et al demonstrated from a sample of 137 participants that the test-retest reliability was 0.91 after a 1-month duration.(Dunst, Trivette & Cross 1986) Sheeran et al found that in a study involving 97 parents of children with epilepsy or cerebral palsy, the internal consistency was 0.87.(Sheeran, Marvin & Pianta 1997) Darling et al found that the internal consistencies for the five subgroups, ranged from 0.48-0.87 when used for the study's population of 120 caregivers to children who had a disability.(Darling & Gallagher 2004)

The validity of the FSS was investigated by Taylor et al, using a population of 990 parents of whom 97% had a child diagnosed with a disability. This study found that

there was a negative correlation between the FSS and stress within a spousal relationship, investigated through the Parenting Stress Index.(Taylor & Others 1993) Hassal et al correlated the FSS to the Parenting Stress Index (PSI) and found an inverse relationship between the two (r=-0.485, p=0.001).(Hassall, Rose & McDonald 2005) Darling et al correlated FSS to family needs and also established that a negative correlation occurred (r=-0.319).(Darling & Gallagher 2004)

4.7.6.2 Family Needs Survey (FNS) (Bailey & Simeonsson 1988)

The Family Needs Survey is a 35-item self-report questionnaire. It was originally developed to assess the functional needs of parents who had young handicapped children.(Bailey & Simeonsson 1988) The needs assessed are: information (7 items), support (8 items), financial (6 items), explaining to others (5 items), community (3 items), child-care (3 items) and professional support (3 items).(Bailey & Simeonsson 1988)

This is an instrument that has been used in assessing needs of parents who have a child with a chronic condition.(Bailey & Simeonsson 1988; Cate, Kennedy & Stevenson 2002) It is an important element to assess as unmet needs have been associated as a source of stress for families, and those caring for a child with a chronic condition display high levels of unmet needs. Farmer et al displayed this by reporting that 93% of the 83 parents asked who had a child with a chronic condition, felt that they had at least one unmet need.(Farmer et al. 2004) It was designed by Bailey et al as a way for parents to express their needs at the time of when a family care plan was to be implemented.(Bailey & Blasco 1990) Therefore, it is an important tool to include within this present study, as it will assess whether parents feel that they have unmet needs as well as facilitating the future aim of the study, which is to provide relevant support for the families that have a child with a potential or new diagnosis of epilepsy.

The scoring of the questionnaire varies across the studies that have used it. Bailey et suggested that a scoring of 1-2-3 should be used, with the highest score indicating that the individual felt that this was a need of theirs.(Bailey & Simeonsson 1988) Other studies who have adopted this scoring have only reported upon the items that were scored as a '3' in order to assess the definite needs felt by the individual who was

completing the questionnaire.(Cate, Kennedy & Stevenson 2002; Farmer et al. 2004) In order to determine definite needs, a scoring of 0-0-1 was employed by Trute et al, allowing for a clear assessment in the areas that the individual felt represented an unmet need.(Trute & Hiebert-Murphy 2005) This was the scoring used by this present study.

The rating of the scale is 0-0-1; 0 indicating that the carer does not need help or is unsure as to whether or not they need help with the item being enquired about, with 1 indicating a definite need for further assistance. Therefore, the higher the score, the more unmet needs being expressed by the carer.(Trute & Hiebert-Murphy 2005)

For the purpose of this study, three areas of need being assessed are of particular interest as they represent the needs that could be provided for within a clinical setting. These are information needs, resource needs and counselling needs. Based upon the study by Trute et al, a 30-item analysis of the FNS will be performed to assess counselling, resource and information needs said to be definitely required by the carer. (Trute & Hiebert-Murphy 2005)

Within the 30-item FNS, 3 items will assess counselling needs, 20 items will assess resource needs and 7 items will assess the carer's information needs. This provides a range of 0-30 for the total number of needs, with the range of scores for each subgroup being as follows: Information (0-7), Resource (0-20) and Counselling (0-3).

High internal consistencies have been demonstrated by previous studies when examining the total FNS score. Sexton et al finding that the internal consistency of the FNS was 0.91 when applied to 53 mothers who had a child with a disability.(Sexton, Burrell & Thompson 1992) Farmer et al also found a high internal consistency, with their study recording the internal consistency to be 0.88.(Farmer et al. 2004) Bailey et al reported that over a 6-month period, the test-retest reliability was 0.67 when given to a sample of 68 parents.(Bailey & Simeonsson 1988)

Trute et al found from their study of 102 mothers that the internal consistencies for the three subgroups being used in this present study were; 0.87 for counselling needs, 0.84 for resource needs and 0.78 for information needs. Test-retest reliability after a 2-week period was also performed using the same subgroups, with the results being 0.90, 0.80 and 0.79 for the respective counselling, resource and information needs.(Trute & Hiebert-Murphy 2005)

4.7.7 Child Self-Image and Self-Esteem

4.7.7.1 Self Image Profiles (SIP)(Butler 2001)

The self-image profile was developed in 2001 by Butler as a brief self-report questionnaire completed by children between the ages of 7-16 years. It provides a visual display of the child's own self image and self esteem.(Butler 2001)

There are two different version of the SIP as it is age dependent. The child version (SIP-C) is completed by those aged between 7-11 years and the adolescent version (SIP-A) is completed by those ages between 12-16 years.(Butler 2001)

There are 25 items, each depicting a self-description, within both forms of the SIP. Butler states that there are 12 items representing a positive self-description, 12 items representing a negative self-description and 1 neutral item in order to reflect 'I'.(Butler 2001)

The positive descriptions produce the 'Positive Self Image' score (SI +ve), the negative descriptions produce the 'Negative Self Image' score (SI –ve) and the score from the neutral item produces the 'Sense of Difference' score.(Butler 2001)

Using the self-image score, different factors can be examined. Butler identified 7 different aspects of self image within the SIP-C and 10 different aspects within the SIP-A.(Butler & Gasson 2005) For the purpose of the analysis, only the SI +ve and SI –ve will be examined to indicate overall self-image rather than looking at individual factors. This will allow for a clearer comparison of self-image differences between the children who have a potential new diagnosis of epilepsy.

The scoring of the SIP is on a Likert scale of 0-6: 0= not at all, 6= very much.

The respondent initially scores each descriptive item according to 'how they would describe themselves'. This produces the SI +ve (0-72), SI –ve (0-72) and SD (0-6) scores (ranges in brackets). A higher SI +ve score reflects the individuals viewing themselves as having more positive attributes. Conversely, a higher SI –ve score reflects the view that they have more negative attributes. The SD score reflects how unique the individual feels that they are, with a higher score representing that individual feeling separate from others.(Butler 2001)

The next stage of completing the SIP is to then rate each descriptive item again, this time referring to 'how they would like to be'. The difference between the initial and second rating then generates the final score, the score reflecting that individual's 'Self

Esteem' (SE). The lower the SE score, the higher the individual's self-esteem as this describes a smaller discrepancy between the two scores.(Butler 2001)

In total, the 25 items are rated twice, first describing their self-image and second describing their self-esteem. A total of four scores are generated.

Butler investigated the internal consistency and the convergent validity of the SIP-C and the SIP-A. The SIP-C was given to 513 children from 5 primary schools in Leeds and found the SI +ve and SI –ve to have a Cronbach's alpha coefficient of 0.69. Construct validity was obtained using the same population group, by comparing the SIP to the Self Perception Profile for Children (SPPC). SE correlation was -0.24, SI +ve was 0.28 and SI –ve was -0.28. (Butler 2001; Butler & Gasson 2005)

The SIP-A was given to 341 adolescents at 3 different secondary schools in Leeds. Internal consistency for SI +ve was 0.69 and SI –ve was 0.79. Again using the same population group, construct validity was obtained by correlating the SIP to the SPPC. SE correlation was -0.28, SI +ve was 0.29 and SI –ve was -0.20. (Butler 2001; Butler & Gasson 2005)

The SIP has so far been used in studies assessing the wellbeing of children who are undergoing anger management,(Down et al. 2011) those who have been identified as being a bully,(Kaloyirou & Lindsay 2008) diagnosed with nocturnal enuresis(Robinson et al. 2003) and those who have undergone a microsurgical transfer of a toe to a hand.(Bellew, Haworth & Kay 2011)

Bulter states that after reviewing 7 alternative scales that assess self-esteem or self-concept, the SIP is the only measure that explicitly frames the descriptive items from the personal construct theory. This was felt to provide the measure with more relevant items to accurately assess a child's self image.(Butler & Gasson 2005) As self-image and self-esteem have been identified as becoming negatively affected within the paediatric epilepsy population,(Hoare & Mann 1994) it was felt to be a relevant measure to include within this study methodology.

4.7.8 Child Behavioural Difficulties

4.7.8.1 Strengths and Difficulties Questionnaire (SDQ) (Goodman 1997, 1999)

Goodman developed the Strengths and Difficulties Questionnaire (SDQ)(Goodman 1997) and an extended version was created in 1999.(Goodman 1999) The SDQ is a brief behavioural screening questionnaire that is completed by the parent, assessing the child's behaviour, emotions and relationships. The proxy report can be completed for children aged 4-16 years old. (Goodman 1997)

The SDQ assesses 25 different attributes, some positive and some negative. From the 25 attributes, 5 different domains were identified.(Goodman 1997, 2001) These are:

- Conduct problems
- Emotional problems
- Hyperactivity-inattention
- Peer Problems
- Prosocial behaviour

Summing the four problem areas creates a total difficulties score: conduct, emotional, hyperactivity and peer problems. The higher the score, the more difficulties are observed.(Goodman 1997)

The extended version used within this study, additionally includes a section to assess the impact that their child's social impairment has upon the home life, friendships, classroom learning and leisure activities. Goodman chose these areas to assess impact as they represent the main domains considered when rating psychosocial disability. (Goodman 1999)

The response for each of the 25 attributes being assessed within the SDQ has three options: 'Not True', 'Somewhat True' or 'Certainly True'.

The scoring for each response depends upon whether the attribute is classified as being positive or a negative:(Goodman 1997)

'Not True' is scored as a '0' if it is a negative attribute, but is scored as a '2' if it is a positive attribute that is being assessed.

'Somewhat True' is always scored as a '1'.

'Certainly True' is scored as a '0' if it is a positive attribute, but it scored as a '2' if it is a negative attribute that is being assessed.

The Impact score is calculated on a scoring system of 0-0-1-2: 0 = not at all/only a little, 1= quite a lot, 2 = a great deal. The higher the score, the more significant the

social impairment impact.(Goodman 1999) The score for each domain can range from 0-10, with the total difficulties score ranging from 0-40.

The SDQ has been demonstrated to display good reliability and validity. A sample of 346 parents completed the SDQ and the Rutter questionnaire. Comparing the total difficulties score of the SDQ to the total Rutter score produced a high correlation of 0.88, indicating good concurrent validity. A Receiver Operating Characteristic (ROC) curve was produced and this displayed an area under the curve being 0.87 for the total difficulties SDQ, the same value achieved using the total deviance score from the Rutter questionnaire. This displayed that the SDQ had predictive validity in being able to determine between a psychiatric and non-psychiatric case.(Goodman 1997)

A larger study involving 9998 parents produced an average Cronbach alpha coefficient across the domains as 0.73. Test-retest stability was also assessed, involving 2091 parents after an interval of 4-6 months with the mean stability being 0.62.(Goodman 2001)

The SDQ has been used as a behavioural screen but also as a means for identifying psychopathology.(Goodman 2001; Goodman et al. 2000) Previous studies involving those diagnosed with epilepsy have used the SDQ when examining child quality of life.(Brunklaus, Dorris & Zuberi 2011; Turky et al. 2008)

A recent study of 152 parents who have a child diagnosed with Dravet Syndrome, used the parent-rated SDQ and found it to be the strongest predictor for health related quality of life within a multivariate regression (beta = -0.43).(Brunklaus, Dorris & Zuberi 2011)

4.7.9 Child Quality of Life

4.7.9.1 The Pediatric Quality of Life Inventory Version 4 (PedsQL 4.0)(Varni, Seid & Kurtin 2001)

Varni et al designed the Pediatric Quality of Life Inventory (PedsQL 4.0) in 2001 as a child self-report questionnaire, measuring health-related quality of life (HRQOL) within the past 1 month. The PedsQL is a generic measure of HRQOL but it was designed to incorporate both generic and disease-specific elements into a singular outcome measurement, making it an appropriate tool to use when a diagnosis of a condition has not been confirmed. It has been validated for children aged 5-18, making it an appropriate measure to use for this study.(Varni, Seid & Kurtin 2001)

The PedsQL 4.0 consists of 23 items, creating 4 domains. They are:

- Physical Functioning (8 items)
- Emotional functioning (5 items)
- Social functioning (5 items)
- School functioning (5 items)

The rating of the PedsQL 4.0 is on a Likert scale of 0-4: 0 = never a problem to 4 = almost always a problem. The scoring of the PedsQL is reversed, with a maximum score being 100 for each item and the minimum being 0. The rating and scoring of each item occurs as: 0=100; 1=75; 2=50; 3=25; 4=0. The scores for each domain as well as the total score are converted so each has a possible range of 0-100, with the highest scores indicating better HRQOL. (Varni, Seid & Kurtin 2001)

Varni et al found the internal consistency of the PedsQL to be 0.88 for the total score with the domains ranging from 0.68-0.83. These values were calculated using 960 children with either an acute or chronic health condition or with no health issues. Construct validity was also determined using the same population group, with healthy children demonstrating a higher PedsQL score than those with a health issue.(Varni, Seid & Kurtin 2001) Another study by Varni et al that involved 2437 school children, also demonstrated that the PedsQL produced higher scores for those that are healthy after controlling confounders (ANCOVA; $F=44.29$, $p<0.001$) with the internal consistency being 0.89 for total score.(Varni, Burwinkle & Seid 2006) PedsQL has been used previously in studies examining HRQOL in those who have epilepsy.(Brunklaus, Dorris & Zuberi 2011; Haneef et al. 2010; Modi et al. 2009; Wood et al. 2008) Haneef et al demonstrated in a study involving 67 children with

epilepsy that the PedsQL demonstrated good internal consistency (0.90) and construct validity by generating a significant correlation between the HRQOL score and the duration of the epilepsy.(Haneef et al. 2010)

4.7.10 Demographic and Fit Information Sheet

The main carer completed a demographic questionnaire to record their race/ethnicity, age, employment status and level of education reached by the main carer. In addition, the carer will provide information regarding the family structure and the suspected epileptic episodes. These include the frequency of the seizures, the length of time that they have been occurring for, their thoughts on what may be happening and whether or not a seizure has occurred in a public place. This will provide a brief overview of the characteristics of the carer that has been enrolled into the study as well as an insight to whether there have been differing experiences of the suspected epileptic episodes.

4.8 Statistical Analysis

Statistical Advisor: Dr Steven Lane, University of Liverpool

The analysis of the results obtained from the different questionnaires was descriptive, with summary statistics of the total score for each questionnaire and the subgroups. Descriptive analysis of the demographics and fit characteristics were additionally performed.

Statistical analysis was undertaken to examine for any associations between carer stress and the investigated carer-completed variables. Initially, it was wanted to assess the relationship between the carer stress, using their average PIP score, and each carer-completed variable, using the total score of the carer-completed questionnaire. As the results could not be assumed as being normally distributed, a Spearman's Rank Correlation was chosen. The next step of the analysis was to determine if there are any differences in variable scores between carers who were categorised, using their average PIP score, as being stressed or not stressed. As the data was non-parametric with small sample sizes for each arm, a Mann Whitney U test was performed to assess for an increased probability of finding a difference in scores between the two groups of carers. The final aim of the statistical analysis was to predict the relationship of a carer being stressed from the variables that had been identified within the Mann Whitney U test as having a significant difference in scores between the carers that are stressed or

not stressed. As it could not be assumed that there was a linear relationship between stress and the variables and as the data was non-parametric, a Binary Regression Model was performed to provide an indication as to which variables are predictive of carer stress.

All questionnaires were compared using the total score. The only exception to total questionnaire score being used was the Brief COPE Inventory whereby the subgroups were used in the analysis.

Due to the small sample size, no further analysis was performed. In addition, no attempt was made to assess for statistical differences between the subgroups of each questionnaire or to assess for associations between the subgroups and carer stress.

5.1 Population Data

Between the period of September 2010 to June 2011, 148 patients were deemed eligible and were sent participation packs.

Of the 148 patients, 133 had a clinic appointment scheduled within the recruitment period and attempts were made to meet them in order to gain consent.

From the 133 carers approached, 60/133 (45%) gave consent for them and their child to partake in the study.

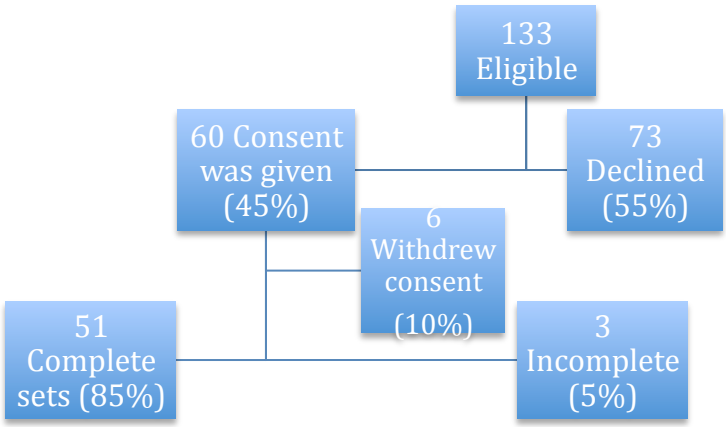
73 carers and children did not give consent. The reasons were:

- 37/73 (15%) were not interested
- 20/73 (27%) did not attend the clinic appointment
- 12/73 (16%) were unable to be met by the researcher
- 4/73 (6%) had scheduled clinic appointments outside of Alder Hey Children's Hospital

Of the 60 carers and their child that said they would like to participate, 6 carers decided to withdraw from the study. This altered the number of carers that consented to be in the study to 54/133 (41%).

Of the 54 participants enrolled into the study, 3 of the questionnaire packs were incomplete due to missing data. This resulted in 54 completed questionnaire packs, with 3 being incomplete. This can be seen diagrammatically in Figure 11.

Figure 11: Schematic Diagram of Recruitment Numbers



In addition to the 54 sets of data, 5 patients' results that were obtained during the pilot study between June 2010 and August 2010 were used in the analysis. It should be noted that these 5 patients' results did not include the semi-structured questionnaire, as this had not yet been passed by ethics during the pilot study.

The end result was that 59 data sets were used for the analysis, with 8 data sets being incomplete. The incomplete data sets were still used in the analysis.

Participant characteristics were given by the main carer and are detailed in Table 14.

Table 14: Carer and Child Demographics

Characteristic	Frequency
Relationship to child (biological, step or foster)	
Mother	54/59 (92%)
Father	3/59 (5%)
Other	2/59 (3%)
Age of carer	
<20	1/59 (2%)
21-29	7/59 (12%)
30-39	34/59 (57%)
40 +	17/59 (29%)
Family structure	
Both parents together (biological/step/foster)	36/59 (61%)
Parent alone	14/59 (24%)
Other	9/59 (15%)
Number of children in the house	
1	12/59 (20%)
2	26/59 (44%)
3	16/59 (27%)
4+	5/59 (9%)
Illness present in other children?	
Yes	12/59 (20%)
No	47/59 (80%)
Employment status of carers	
Employed	41/59 (69%)
Unemployed	18/59 (31%)
Level of education of carer	
Left school at 16	24/59 (41%)
Left school at 18- no further education	9/59 (15%)
Left school at 18- went onto further education	26/59 (44%)
Gender of child	
Female	28/59 (47%)
Male	31/59 (53%)
Age of Child	
0-6 years	24/59 (41%)
7-11 years	18/59 (30%)
12-16 years	17/59 (29%)

The carer was additionally asked about the fit that resulted in their child being referred with a possible diagnosis of epilepsy. It was enquired as to whether it had occurred within a public place, how many times the fit had occurred and over what duration of time. An additional question asked as to what the carer thought was the underlying cause of the fit, assessing whether the carer viewed the fit as being epilepsy or not. The results can be seen in Table 15.

Table 15: Fit Characteristics as given by the Carer

Characteristic	Frequency
Have they had a fit in a public place?	
Yes	41/59 (69%)
No	18/59 (31%)
Fit frequency since they have been occurring	
1-4 x total	23/59 (39%)
1-2 x a month	6/59 (10%)
1-2 x a week	8/59 (14%)
3-5 x a week	7/59 (12%)
Everyday	15/59 (25%)
Length of time fits have been occurring	
1-2 months	8/59 (13%)
3-4 months	11/59 (19%)
5-6 months	6/59 (10%)
7+ months	34/59 (58%)
Carers thoughts as to what the problem is	
Epilepsy	27/59 (46%)
Not sure	15/59 (25%)
Normal	9/59 (15%)
Other diagnosis	8/59 (14%)

5.2.1 Questionnaires Examining Carer Stress

5.2.1.1 Pediatric Inventory for Parents (PIP) Total Score Results

A total of 56 carers completed the PIP, with the median value for the PIP-frequency score being 96 and the PIP-difficulty being 90. Descriptive statistics for the PIP can be seen in Table 16 with the frequency of the score obtained in each domain shown in Figure 12 and Figure 13.

Using the pre-determined cut-off score of 90, 43% of the carers were categorised as being stressed. See methodology for reasoning behind the decided cut-off value.

Table 16: Summary Statistics of Total PIP Score Results

	Number of carers	Mean	Median	Mode	Std. Deviation	Range of scores	Inter-quartile range
Total PIP-d	56	93.11	90.00	122	29.58	46-174	68-109
Total PIP-f	56	95.54	96.00	56	25.76	56-154	72-114

Figure 12: PIP Difficulty Score Histogram

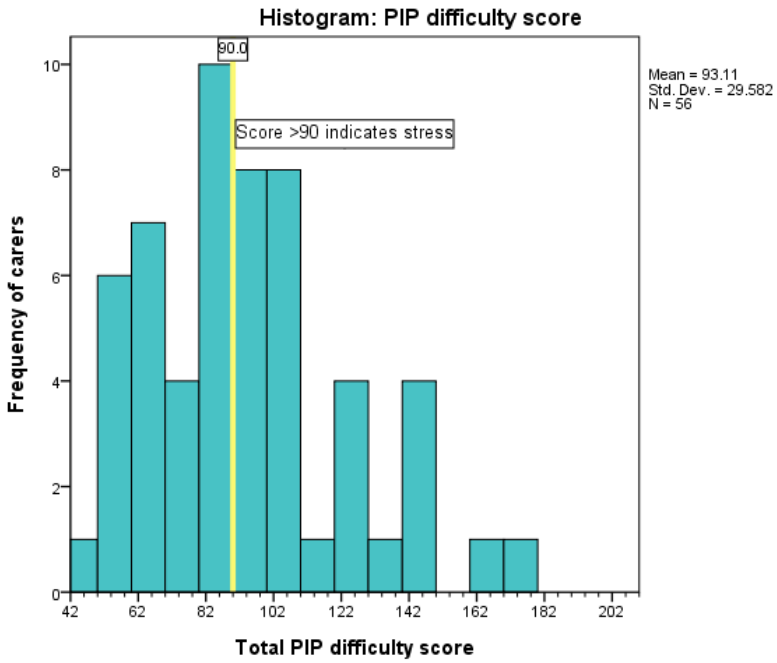
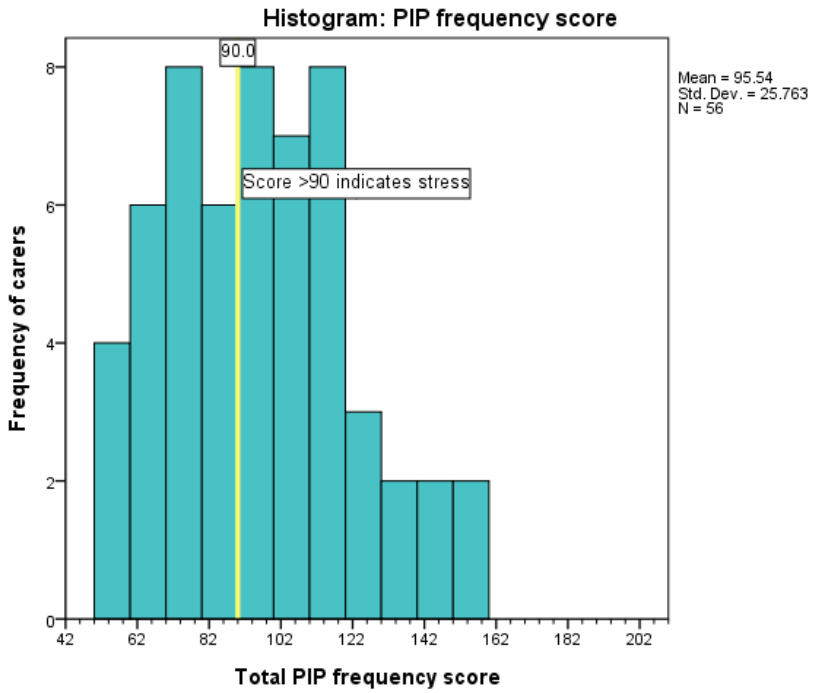


Figure 13: PIP Frequency Score Histogram



6.2.1.2 Semi-Structured Stress Questionnaire-Quantitative Results

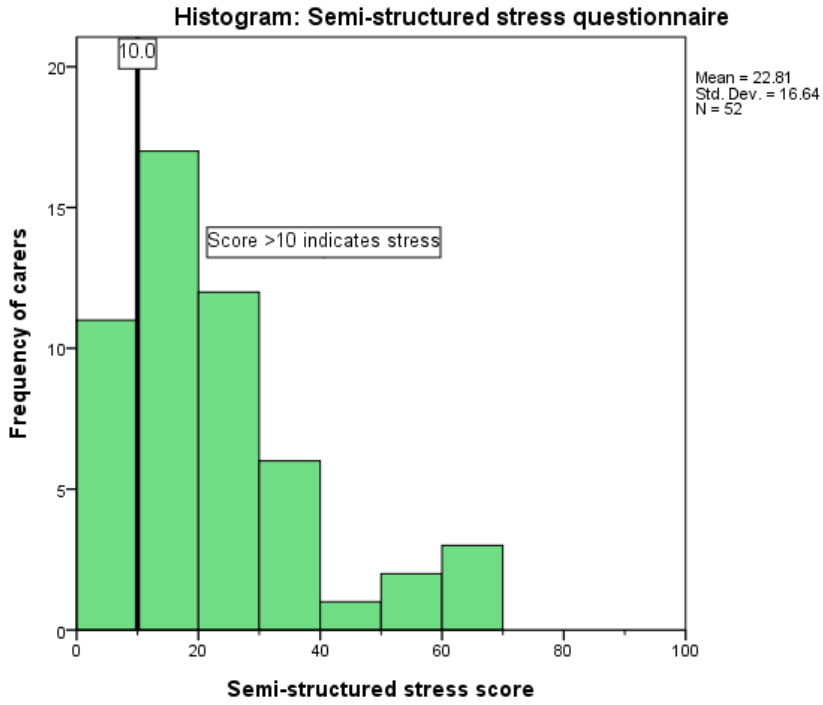
A pre-determined score of 10 was used as a cut-off value so to indicate whether or not a carer was experiencing significant stress. The total number of carers that completed the semi-structured stress score was 52. The mean semi-structured stress score for this population was 22. Descriptive statistics of the total score for the semi-structured stress questionnaire can be seen in Table 17.

Table 17: Summary of Semi-Structured Stress Questionnaire Quantitative Results

	Number of carers	Mean	Median	Mode	Std. Deviation	Range of scores	Inter-quartile range
Semi-structured stress score	52	22.81	18.00	18	16.64	0-69	10-29

The range of scores along with the cut-off value used for this questionnaire can be seen in Figure 14.

Figure 14: Semi-Structured Stress Questionnaire Quantitative Results Histogram



5.2.2 Questionnaires Examining Carer Resources

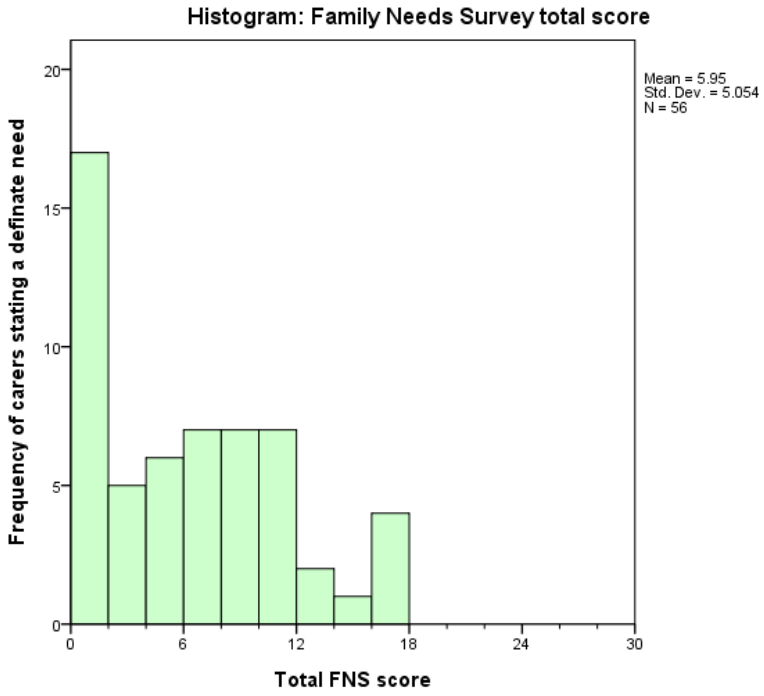
5.2.2.1 Family Needs Survey (FNS) Results

A total of 56 carers completed the FNS. The scoring of the FNS was 0-0-1, enabling the needs that were definitely required to be surveyed. The mean number of needs definitely wanted was 5.95 per carer. The most frequently reported number of needs was 0 although the range of needs reported was wide, with the maximum number being 17 needs were required. The descriptive statistics can be seen in Table 18 with Figure 15 displaying a histogram for the total FNS score number.

Table 18: Summary Statistics of the FNS

	Number of carers	Mean	Median	Mode	Std. Deviation	Range of scores	Number of items	Standardised mean
Total FNS	56	5.95	5.50	0	5.05	0-17	30	-
Resource needs	56	3.04	2.00	0	3.35	0-12	20	0.125
Information needs	56	2.66	3.00	0	2.11	0-7	7	0.38
Counselling needs	56	0.25	0	0	0.61	0-3	3	0.08

Figure 15: FNS Total Score Histogram



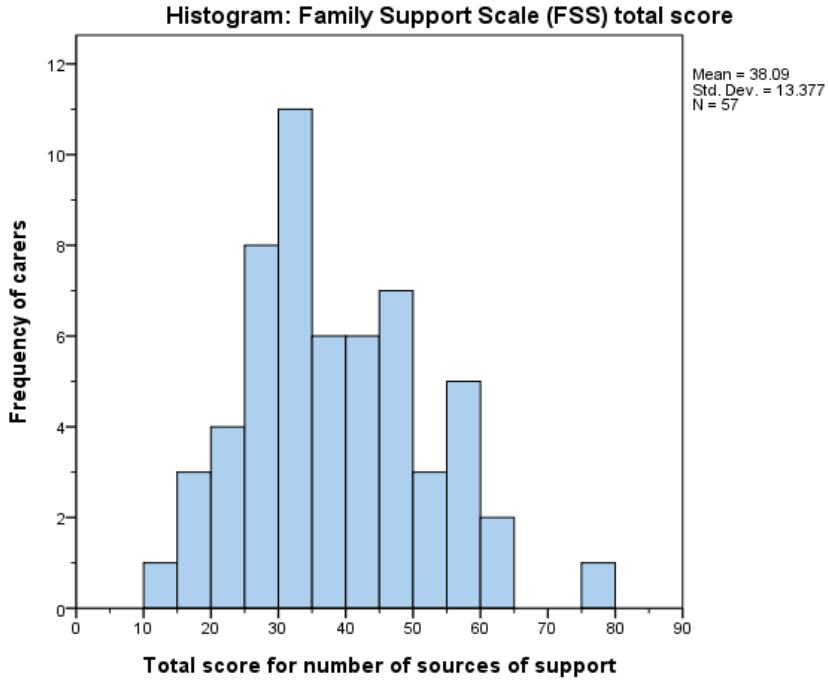
5.2.2.2 Results from the Family Support Scale (FSS)

A total of 57 carers completed the FSS. The total score for the FSS can range between 0-90. This population reported a range of available sources of support to range between 14-78, with the mean being 38.09 sources of support reported to being available to the carer. There were multiple modals for number of sources of support with 27 sources being the smallest reported mode. Descriptive statistics for the FSS can be seen in Table 19 with a histogram for the total FSS score displayed in Figure 16.

Table 19: Summary Statistics of the FSS

	Number of carers	Mean	Median	Mode	Std. Deviation	Range of scores	Maximum score possible	Standardised mean
Total FSS	57	38.09	37.00	27	13.38	14-78	90	-
Partner support	57	8.04	8.00	8	4.5	0-15	15	0.536
Informal support	57	10.93	10.00	8	5.43	0-24	25	0.437
Formal support	57	5.86	6.00	5	2.86	0-10	10	0.586
Social organisation	57	6.28	6.00	5	3.75	0-19	20	0.314
Professional services	57	6.98	7.00	7	2.55	3-12	20	0.349

Figure 16: FSS Total Score Histogram



5.2.3 Questionnaire Examining Carer Coping Strategies

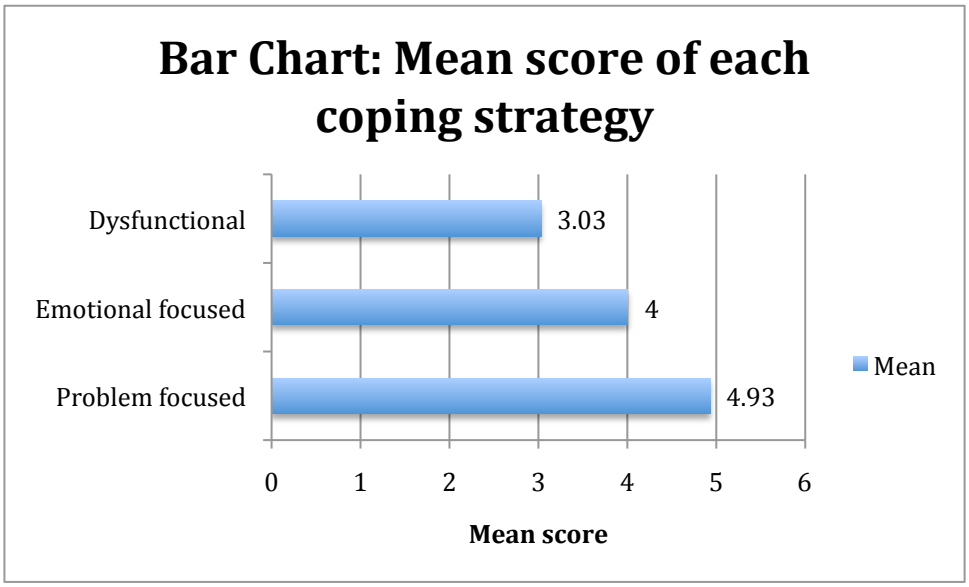
5.2.3.1 Brief COPE Inventory Results

A total of 55 carers completed the Brief COPE Inventory and this was the only questionnaire used that did not generate a total score. The reason being that too much information was lost as the purpose of the questionnaire was to indicate the type of coping strategies employed rather than the total number. For this reason, the questionnaire will present the subgroups that were created. Descriptive statistics can be seen in Table 20 with Figure 17 displaying the mean score for each coping strategy.

Table 20: Summary Statistics of the Brief COPE Inventory

	Number of carers	Mean	Median	Mode	Std. Deviation	Range of scores	Items in each group	Standardised mean
Problem focused	55	14.78	15.00	12	4.89	6-24	3	4.93
Emotional	55	20.00	20.00	19	5.23	10-35	5	4.00
Dysfunctional	55	18.20	17.00	12	5.69	12-34	6	3.03

Figure 17: Mean score for each Coping Strategy Bar Chart



5.2.4 Questionnaire Examining Carer Locus of Control

5.2.4.1 Internal-External Locus of Control Results

A total of 55 carers completed the Internal-External Locus of control. The range of possible scores for this questionnaire is 0-23. This study found that the range of reported scores was 6-22. Descriptive statistics can be seen in Table 21 with Figure 18 displaying the frequency of each score and Figure 19 displaying the mean score for each locus of control category.

Table 21: Summary Statistics of the Internal-External Locus of Control

	Number of carers	Mean	Median	Mode	Std. Deviation	Range of scores
Total score	55	12.33	12.00	10	3.79	6-22

Figure 18: Locus of Control Score Histogram

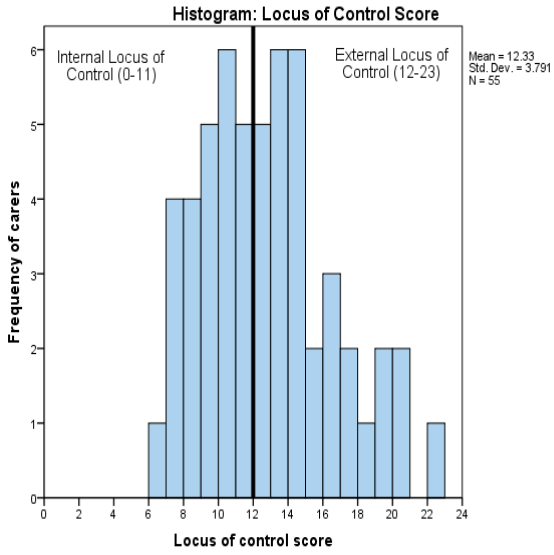
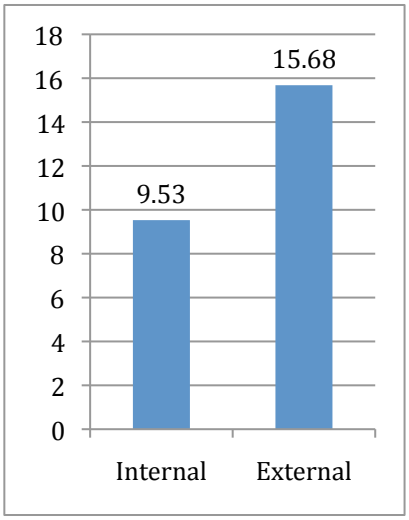


Figure 19: Mean score for each locus of control



Based on previous studies, (Parkes 1984; Renn & Vandenberg 1991) the median value was used so to categorise the carers as having an internal or an external locus of control. The median was found for this sample to be 12. This can be seen in table 21. Therefore scores from 0-11 formed the internal group and 12-23 formed the external group. Figure x displays the frequency of the locus of control scores, with figure y displaying the mean score for each categorisatory of either external or internal locus of control.

5.2.5 Questionnaire Examining Carer Health

5.2.5.1 General Health Questionnaire-28 (GHQ-28) Results

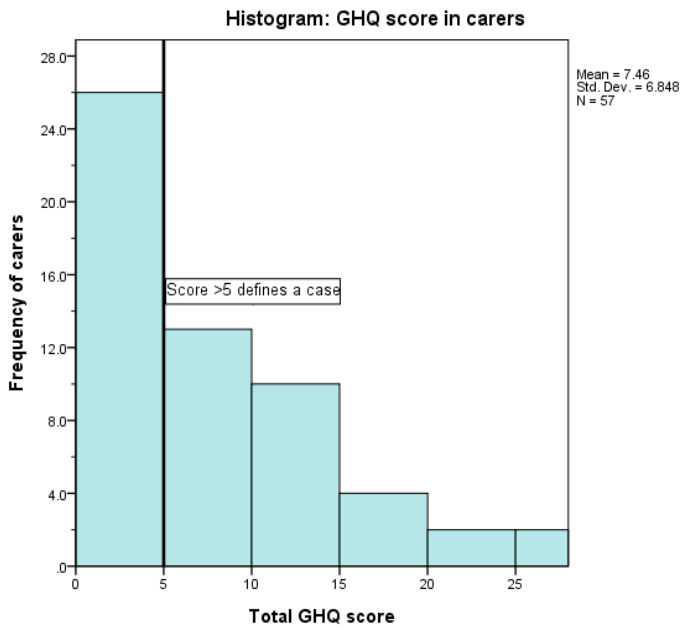
A total of 57 carers completed the GHQ-28. The GHQ-28 was scored using the GHQ scoring system and therefore a maximum number for each of the four subscales was 7, making the maximum total score possible being 28.

A threshold of 5 was suggested by Goldberg et al to be used to indicate ill mental health.(Goldberg et al. 1997) This threshold limit can be seen in Figure 20 that displays the frequency for the GHQ-28 total score. Table 22 displays the descriptive statistics for the questionnaire.

Table 22: Summary Statistics of the GHQ-28

	Number of carers	Mean	Median	Mode	Std. Deviation	Range of scores achieved	Inter-quartile range
Total GHQ-28	57	7.46	6.00	0	6.85	0-26	2-13
Somatic symptoms	57	2.54	2.00	0	2.24	0-7	1-5
Anxiety and Insomnia	57	2.89	3.00	0	2.37	0-7	0-5
Social Dysfunction	57	1.28	0.00	0	1.88	0-6	0-2
Depression	57	0.74	0.00	0	1.68	0-7	0-0

Figure 20: GHQ Total Score Histogram



5.2.6 Questionnaire Examining Family Functioning

5.2.6.1 Family Adaptability and Cohesive Evaluation Scale-IV (FACES-IV)

Results

A total of 54 carers completed FACES-IV. The areas that this thesis was most interested in were the total circumplex ratio, the family satisfaction and the family communication. The later two scales are reported as a percentage as this is what FACES-IV administration pack recommends.(Olson, Gorall & Tiesel 2006) The summary statistics can be seen in Table 23.

The circumplex ratio provides a measure of the level of balance versus unbalance in a system and therefore is a good overview measure of family functioning. A ratio score of 1 displays equal balance and any ratio above 1 shows a more balanced or healthy family system. Olson states that most families will have a total ratio between 0-2.(Olson 2011) The histogram of the total circumplex ratio scores can be seen in Figure 21, with Figure 22 and Figure 23 displaying the results for family satisfaction and communication.

Table 23: Summary Statistics of FACES-IV

	Number of carers	Mean	Median	Mode	Std. Deviation	Range of scores
Total Circumplex ratio	54	1.96	2.06	2.06	0.61	0.63-3.19
Family satisfaction (%)	54	53.76	58.00	10	30.92	10-99
Family communication (%)	54	64.17	70.00	24	29.10	10-99

Figure 21: Total Circumplex Ratio Score Histogram

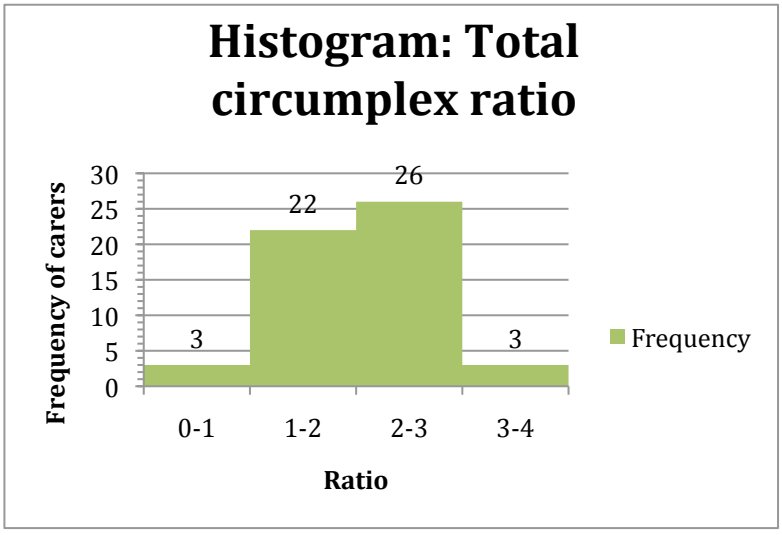


Figure 22: Family Satisfaction Scale Scores Histogram

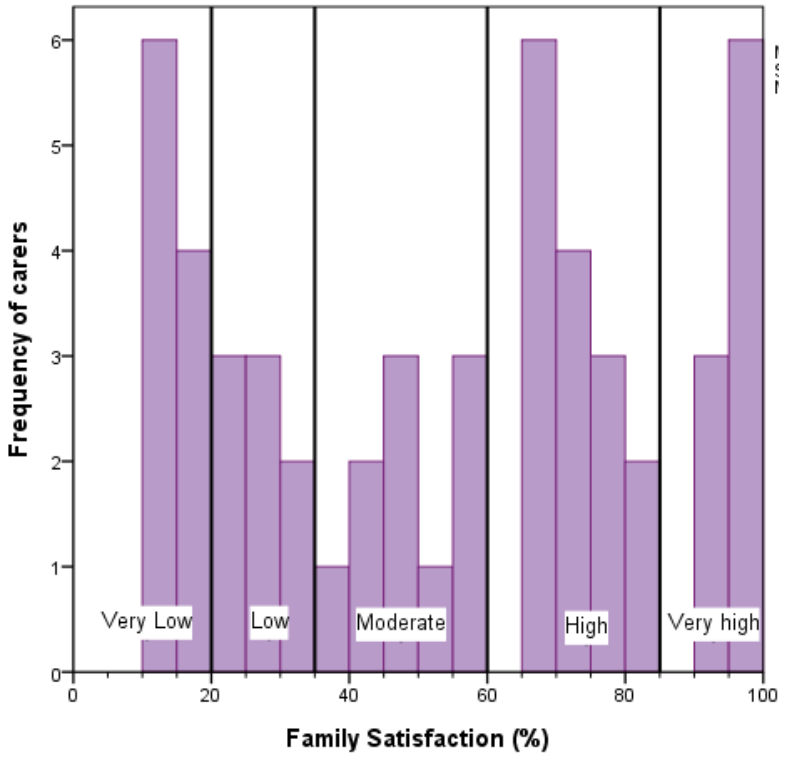
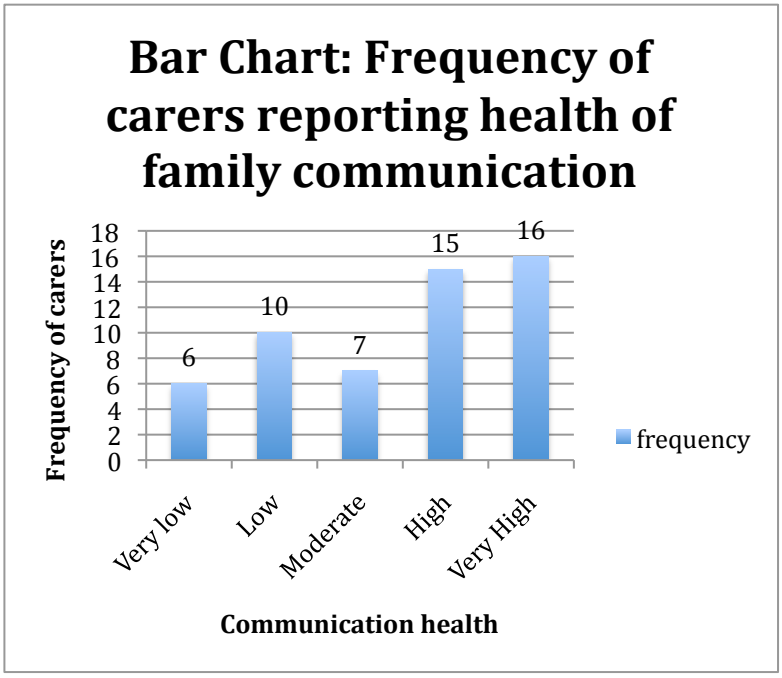


Figure 23: Family Communication Scale Scores Bar Chart



5.2.7 Questionnaire Examining Child Behavioural Difficulties

5.2.7.1 Strengths and Difficulties Questionnaire (SDQ) Results

A total of 51 carers completed the SDQ. Table 23 displays the descriptive statistics. The cut-off values for determining whether the child's difficulties are within the normal, borderline or abnormal range are shown in the last 3 columns of Table 24. These were taken from the SDQ administration information.(Youth In Mind 2011) Table 25 and Figure 24 displays the frequency of children within each category.

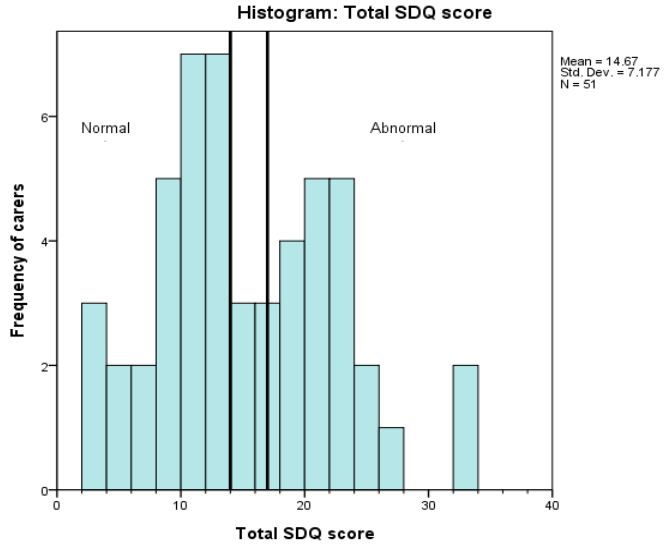
Table 24: Summary Statistics of the SDQ

	Number of carers	Mean	Median	Mode	Std. Deviation	Range of scores	Normal range	Border-line	Abnormal
Total SDQ score	51	14.67	13.00	10	7.18	2-32	0-13	14-16	17-40
Emotional problems	51	3.57	3.00	1	2.74	0-10	0-3	4	5-10
Conduct problems	51	2.65	3.00	1	1.72	0-8	0-2	3	4-10
Hyperactivity	51	5.86	6.00	5	2.86	0-10	0-5	6	7-10
Peer problems	51	2.59	2.00	0	2.26	0-9	0-2	3	4-10
Prosocial	51	7.69	8.00	10	2.45	0-10	6-10	5	0-4
Impact score	51	1.73	1.00	0	2.30	0-8	0	1	2-8

Table 25: Categorisation of Behavioural Difficulties

	Number of patients in each category (N=51)		
Subgroups within the SDQ	**Normal**	**Borderline**	**Abnormal**
Total SDQ score	26	5	20

Figure 24: SDQ Total Score Histogram



5.2.8 Questionnaire Examining Child Quality of Life

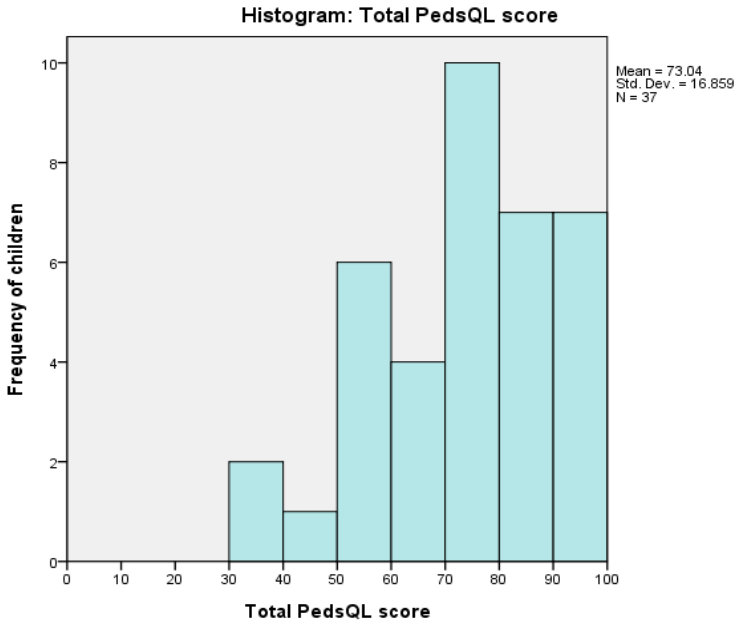
5.2.8.1 Paediatric Quality of Life 4.0 (PedsQL 4.0) Results

A total of 37 children completed the PedsQL 4.0. The normal scores are the average scores taken from a healthy sample of 5480 children.(Varni, Limbers & Burwinkle 2007) Descriptive statistics can be seen in Table 26, with Figure 25 displaying a histogram for the total score.

Table 26: Summary Statistics of the PedsQL 4.0

	Number of children	Mean	Median	Mode	Std. Deviation (SD)	Range of scores	Normal mean score (SD)
Total PedsQL	37	73.04	78.26	78.26	16.86	30.43-94.57	83.84 (12.65)
Physical	37	80.92	84.38	62.50	16.21	37.50-100	87.53 (13.50)
Emotional	37	66.08	70.00	65.00	25.03	0-100	79.33 (18.15)
Social	37	77.30	80.00	100.00	21.97	20-100	85.15 (16.76)
School	37	60.54	70.00	70.00	22.94	5-90	81.12 (16.45)

Figure 25: PedsQL 4.0 Total Score Histogram



5.2.9 Questionnaire Examining Child Self-Image and Self-Esteem

5.2.9.1 Self Image Profile Questionnaire (SIP) Results

A total of 35 children and adolescents were able to complete the SIP. Of the 35 that completed the questionnaire, 18 were children (7-11) and 17 were adolescents (12-16). The summary statistics of the total population can be seen below in Table 27.

Table 27: Summary Statistics of the SIP Results

	SI +ve	**SI -ve**	**SE**	**SD**
Number of children/adolescents	35	35	35	35
Mean	53.71	28.74	8.46	2.94
Median	55.00	30.00	6.00	3.00
Mode	49.00	12.00	0	6.00
Standard deviation	10.83	15.05	9.48	2.27
Range	29.00-72.00	0-55.00	0-43.00	0-6.00

5.3 Statistical Analysis

5.3.1 Spearman Rank Correlation

The analysis of the individual questionnaires showed that there was variation in the total scores as reported by the carer. As one of the aims of this study is to assess for an association between variable and stress, it was felt necessary to initially perform a correlation between the stress score and the total scores of the individual questionnaires. This thesis is interested in understanding carer stress and the relationship with other variables. Thus, only questionnaires that were completed by the carer will be used in the correlation.

The PIP questionnaire will be used as a representation of the stress scores. This was chosen as the semi-structured stress questionnaire has not been used in published data, whilst the PIP is a stress questionnaire that has been previously validated and used as a measure of stress in other studies, see Table 11. It was therefore felt that the PIP provided the most valid representation of carer stress. The PIP generated two total scores, therefore, an average of the two scores was created for each carer that completed the questionnaire.

The analysis of the individual questionnaires showed that a normal distribution could not be assumed. Therefore, a Spearman Rank correlation was decided upon as it assumes a non-parametric distribution. A two-tail significance was decided as being most appropriate so to not assume a direction of probability.

The results of the Spearman Rank correlation can be seen below in Table 28, displaying the correlation coefficient and the significance of each value.

Table 28: Spearman Rank Correlation between Average PIP Score and the Total Score of Each Questionnaire

	GHQ	SDQ	FSS	Locus	FNS	Dys-cope	Prob-cope	Emo-cope	TCR	F-sat	F-comm
Correlation coefficient	.460	.544	-.152	.184	.403	.392	.344	.320	-.115	-.198	-.111
Sig (2-tail)	.000	.000	.262	.180	.002	.003	.010	.017	.406	.151	.422
N	56	56	50	55	56	55	55	55	54	54	54

Dys Cope= Dysfunctional Coping Strategies; Prob-cope= Problem-focused Coping Strategies;
Emo-cope= Emotional-focused Coping Strategies; TCR= Total Circumplex Ratio;
F-sat= Family Satisfaction Scale; F-comm= Family Communication Scale.

The different colours correspond to the significance of the correlation coefficients. Those coloured red indicate that $p<0.01$ and those coloured blue indicate $p<0.05$.

The strongest correlation was seen between the average PIP score and the SDQ score, $r=0.544$, $p<0.01$. From these results, it appears that the strongest association was between carer stress and the behaviour difficulties in a child, with more stress being seen as behaviour difficulties increase.

The next two strongest correlations were found between PIP and GHQ ($r=0.460$) and FNS ($r=0.403$). Both of these correlations were positive with a significance of $p<0.01$. However, the coefficient is still reasonably low, suggesting that there is a correlation but the association between increased carer stress and carer health or unmet needs are quite weak.

The three subgroups of the Brief COPE showed that the dysfunctional coping strategy ($r=0.392$, $p<0.01$) was the most associated to increased carer stress out of the three coping strategies that were measured. Problem-focused and emotional-focused coping strategies produced similar correlation coefficients but with a lower significance level $p<0.05$. This implies that there may be an association between more dysfunctional coping strategies being used and increased carer stress. However, the coefficient was low, intimating that the association is weak. The Spearman rank identified no correlation existed between stress scores and the total number of sources of support nor the locus of control.

Negative correlations were found between the average PIP score and the FSS, FACES total circumplex ratio (TCR), FACES family satisfaction (F-sat) and FACES family communication (F-comm). This trend in correlation suggests that as the number of sources of support available increases or the family functioning becomes more balanced with more satisfaction with the family and quality of communication, carer stress decreases.

However, all correlation coefficients were below 0.6, which indicates that the correlation being observed may not carry much meaning as ± 0.6 is considered the cut-off value for a correlation to be considered a significant association (Conversation with statistician Dr Steven Lane). Therefore, a Mann Whitney U test will be performed to assess whether there are differences in scores between the carers that are classified as being stressed and those that are not.

5.3.2 Mann Whitney U Test

The Spearman Rank correlation suggested that there are some variables that may be associated with increased carer stress, notably, child behaviour, carer health and unmet needs. It was then decided to compare the total scores of the questionnaire that the carer completed between those that were categorised as being stressed or not stressed. As a normal distribution could not be assumed, a Mann Whitney U test was performed. Categorisation occurred using the average PIP score. A pre-determined cut-off value of 90 was decided upon, based on the mean of previous PIP scores found by studies that have used this measurement. Therefore, carers whose average PIP scores were <90, were classified as being not stressed, whilst those with scores >90 were classified as being stressed. This can be seen in Table 29.

Table 29: Categorisation of Carers Being Stressed

	Frequency	Percentage (%)
Stressed (>90)	32	43
Not stressed (<90)	24	57

Using this categorisation, a Mann Whitney U Test was generated to indicate whether there are any significant differences between the stressed and not stressed groups when comparing the scores of the other variables that were measured via questionnaires. The results of the Mann Whitney U Test can be seen in Table 30.

The null hypothesis was that there is no difference between the questionnaire scores for those that are stressed and not stressed.

Table 30: Mann Whitney U Test Results

Hypothesis Test Summary

	Null Hypothesis	Test	Sig.	Decision
1	The distribution of GHQ is the same across categories of Stressed.	Independent-Samples Mann-Whitney U Test	.008	Reject the null hypothesis.
2	The distribution of SDQ is the same across categories of Stressed.	Independent-Samples Mann-Whitney U Test	.003	Reject the null hypothesis.
3	The distribution of FSS is the same across categories of Stressed.	Independent-Samples Mann-Whitney U Test	.205	Retain the null hypothesis.
4	The distribution of Locus is the same across categories of Stressed.	Independent-Samples Mann-Whitney U Test	.117	Retain the null hypothesis.
5	The distribution of FNS is the same across categories of Stressed.	Independent-Samples Mann-Whitney U Test	.004	Reject the null hypothesis.
6	The distribution of dysfuntion_cope is the same across categories of Stressed.	Independent-Samples Mann-Whitney U Test	.001	Reject the null hypothesis.
7	The distribution of problem_focused_cope is the same across categories of Stressed.	Independent-Samples Mann-Whitney U Test	.054	Retain the null hypothesis.
8	The distribution of emotional_cope is the same across categories of Stressed.	Independent-Samples Mann-Whitney U Test	.020	Reject the null hypothesis.
9	The distribution of FACES is the same across categories of Stressed.	Independent-Samples Mann-Whitney U Test	.986	Retain the null hypothesis.
10	The distribution of fam_sat_per is the same across categories of Stressed.	Independent-Samples Mann-Whitney U Test	.166	Retain the null hypothesis.
11	The distribution of fam_comm_per is the same across categories of Stressed.	Independent-Samples Mann-Whitney U Test	.773	Retain the null hypothesis.

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

Using a significance level of 0.05, 4 questionnaires reported significant differences between carers that are stressed and those that are not. The questionnaires that indicated there was a difference were: General Health Questionnaire (GHQ-28), Strength and Difficulty questionnaire (SDQ), Family Needs Survey (FNS), Brief COPE Inventory subgroups emotional coping and dysfunctional coping. This indicates that these four questionnaires produce significantly different results depending upon whether or not the carer is stressed. These findings agree with the Spearman Rank correlation; that there may be an association between the scores of these questionnaires and the amount of stress being experienced by the carer.

The Mann Whitney U test identified that the most significant difference was with the dysfunctional coping strategy, closely followed by the SDQ and FNS. The findings from the Spearman Rank correlation along with the Mann Whitney U test, indicates that the questionnaires that will be of most use for future interventions in order to predict whether or not a carer is likely to be stressed, are the GHQ, FNS, SDQ and Brief COPE Inventory.

6.3.3 Binary Logistic Regression

In order to determine whether or not these questionnaires can be use in predicting carer stress, a binary logistic regression model was generated using the questionnaires that were significant within the Mann Whitney U test.

Using a binary logistic regression model, the dysfunctional coping strategy when combined with the total SDQ score, both remained significant (0.025 and 0.022 respectively). However, when the FNS total score was added into the formula, only the SDQ remained significant (0.035). This suggests that both the dysfunctional cope and the FNS variables are explaining similar variability in whether or not stress is being experienced by carers.

Therefore, two predictive models were found. When the dysfunctional coping variable was removed, the SDQ and FNS variables were added with either emotional coping strategy or with the GHQ variable. In both cases, the SDQ and FNS remained significant with the other two variables becoming insignificant. The similar findings were found when FNS was removed, and SDQ and dysfunctional coping were left in the equation. When the SDQ and dysfunctional coping variables were added with either emotional coping or the GHQ variables, only the SDQ and dysfunctional coping strategy remained significant. All workings can be seen in Appendix 4.

The binary regression suggests that a combination of either SDQ and dysfunctional coping variables or SDQ and FNS variables are most predictive in determining whether or not a carer is likely to be stressed. This suggests that the best model is based on SDQ and either FNS or dysfunctional coping. This is important to note as for future interventions, it appears that only the SDQ and one of the other questionnaires need to be given to the carer in order to predict carer stress.

Chapter Six

Discussion and Conclusion

6.1 Demographic and Fit Characteristic Discussion

Of the carers, 95% were Caucasian and 92% of the carers were the maternal caregivers. The majority of the caregivers were aged between 30-39 years and lived with another main caregiver. Most carers were employed and had another child living in the house other than the child who was enrolled into the study. Of the children enrolled in the study, the distribution of genders was equal with slightly more males (53%). It was found that only 63% of the children enrolled were old enough to complete one of the questionnaires and 59% were old enough to complete both of the questionnaires. This highlights that there will be a lack of power for the children's questionnaires compared by those completed by the carer.

When asked about the fits that have been occurring, the majority said that they have occurred in a public place. Despite all children presenting to outpatients department with a potential diagnosis of epilepsy, there was a wide range of reported frequencies and length of time that the fits have been occurring for. A surprising number of carers reported that the fits have been occurring for over 7 months before being seen at Alder Hey Children's Hospital. Interestingly, 54% of the carers asked did not think that the fits were occurring due to their child having epilepsy but thought that it was due to another disease process. It was also found that 15% of the carers did not think that their child had had a fit due to viewing the queried seizure, mainly absence episodes, as a normal part of their child's development or behaviour. This may help to explain why high stress levels are not seen throughout all the carers, as there is a lack of acknowledgement of the stressor event. It may also be indicating that carers use a particular type of coping response, which could arguably be denial.

6.2 Interpretation of Questionnaire Results

6.2.1 Discussion of the PIP Results

Both Figure 12 and Figure 13 display that the populations for the PIP scores are not normally distributed. The PIP, for each domain, produces a possible range of 42-210 for the total score. These histograms, presented within Figure 12 and 13, show that the range of scores was wider for the difficulty of the illness-related causes of stress, as perceived by the carer, rather than the frequency. This implies that carers reported a wider range of differing opinions as to how difficult the illness-related stress has been for them, with the smaller frequency range suggesting that carers were more likely to agree with one another as to how often they have been occurring. Using a pre-determined cut-off score of 90, 43% of the carers were stressed. For the carers displaying a different score in the frequency and difficulty domains, an average of the score was taken. This then allowed the carer to be categorised as being stressed if their average PIP score was over 90.

The higher PIP-f mean and median score suggests that carers found the frequency of the events contributed more to the overall stress levels than the perceived difficulty posed by the event. The median score for PIP-d and PIP-f were found to be 90 and 96 respectively. This implies that at least 50% of the carers involved in the study achieved a PIP score that indicated that they were stressed. Of note, the PIP-f scores achieved a higher median score, suggesting that the frequency of the health-related events contributed more to their stress surrounding the possible diagnosis of epilepsy than their perception of how difficult they have been in occurring. The same finding was demonstrated in the mean scores for PIP-d and PIP-f, being 93.11 and 95.54 respectively. In order to gauge how the PIP-d and PIP-f scores in this study compared to previous studies, the mean scores of each domain were used, as this was the only data presented within the other 7 studies, see Table 11.

Looking at the previous scores obtained in the other studies that have used the PIP, presented within Table 11, the PIP-f mean score in this study, 95.54, was found to be higher than mean frequency stress scores in parents whose child has been diagnosed with cancer, diabetes, bladder exstrophy, epilepsy and inflammatory bowel disease. The mean PIP-d score obtained in this study, 93.11, was found to be higher than parents whose child has been diagnosed with sickle cell disease, diabetes, obesity, bladder exstrophy, epilepsy and inflammatory bowel disease. The comparison of the

results obtained in this study compared to previous studies does suggest that the carer population were experiencing more stress than previously identified in other studies. Wagner et al found that 9 parents who have a child diagnosed with epilepsy, reported the mean PIP-f and PIP-d to be 86.1 and 77.6 respectively.(Wagner et al. 2010) The PIP scores obtained in this present sample produced much higher PIP scores, especially for the difficulty domain. However, the sample size by Wagner was a lot smaller than this present sample size and could explain why the scores were a lot lower than this study.

The reason for the higher stress scores found in both domains for this study than demonstrated in previous studies may be due to all other conditions having been diagnosed, whilst the carer population in this study have not yet received a diagnosis for their child. Using this premise, it may be the uncertainty of the diagnosis that may be producing the higher stress scores, especially compared to the previous epilepsy study that used the PIP. The uncertainty of the diagnosis has been shown to be a cause of stress to parents and this may be the reason as to why carers in this study appear to be reporting higher stress levels than found in previous studies.(Lenhard et al. 2005)

When analysing the PIP results, it was important to determine how many carers had been potentially misclassified as being either stressed or not stressed due to only one domain score being above 90. It was found that 6 carers were classified as being stressed due to having an average >90 although one domain score was under this cut-off value. It was additionally found that 2 carers were not classified as being stressed although one domain score was above 90. This means that 8/56 (14%) carers only scored above 90 in one of the domains. It could be suggested that a score above 90 in either domain signifies that a carer is feeling stressed regardless of whether this is due to their perception of the frequency or difficulty of the health-related events. Using this premise, it is reasonable to suggest that 2 of the carers were misclassified as not being stressed despite one score indicating that they were, making this a limitation of the results.

6.2.2 Discussion of the Semi-Structured Stress Questionnaire- Quantitative Results

Figure 14, representing the total scores of the semi-structured questionnaire, displays the wide range of scores reported by the carers to be between 0-69. No carer reported the maximum stress score, a score of 100. The mean stress score was found to be 22.81, with the modal and median value being 18. This, along with the distribution of scores as seen in Figure 14, suggests that the majority of the carers reported a stress score between 0-30. A pre-determined cut-off value as used. This was decided upon as the FBII, the questionnaire that the semi-structured stress questionnaire was based on, used a cut-off value of 10 to indicate stress in parents. This information was gathered from personal communication between Andrew Curran and Jerry Taylor, the creator of the FBII (see methodology for full explanation). In fact, using the cut-off value of 10, 39/52 (75%) of the carers were categorised as being stressed. This is a much higher estimation of carer stress as compared to the PIP, only 43% of the carers are categorised as stressed. The realisation of the discrepancy between categorisation of being stressed as indicated by the two stress questionnaires might suggest that a cut-off value of 10 for the semi-structured stress questionnaire, over-estimates the number of carers that are stressed. This was suggested due to the semi-structured stress questionnaire not having been used before in published data and with the cut-off value of 10 being determined from a group of carers whose child had not been diagnosed with epilepsy.

As a cut-off value has not been firmly established in previous data and nor had this questionnaire been used for parents of children with epilepsy, it was investigated for future guidance on this use of the questionnaire, if an increase in the cut-off value from 10 to 20, was a more appropriate value for carers of children with newly diagnosed epilepsy. Using a cut-off value of 20, this estimated that 24/52 (46%) of the carers were stressed and this agreed much more with the categorisation generated by the PIP. If should be noted that a clinical threshold value for the PIP has not been previously stated. However, the similar percentage of carers being categorised as stressed using a PIP score of 90 and a semi-structured score of 20, suggests that for future work, a score of 20 is a more accurate means for categorising carer stress.

6.2.3 Discussion of the FNS Results

The total score for the FNS represents the total number of needs that were declared by the carer as definitely required. The maximum number that a carer could state was 30, however for this sample of population the maximum number of needs expressed were 17. Despite not being the maximum number of unmet needs possible, 17 unmet needs do show that a lot of carers within this population felt that more help would be of use to them. 77% of the carers who were asked stated that they have at least one unmet need and this was displayed by the mean number of needs for each carer being 5.95, indicating that overall an increase in the amount of help being offered to carers would be of benefit.

The most frequently expressed number of needs was that none were needed, with 13 of the carers stating this fact. This finding supports the notion that the majority of carers do have unmet needs although not all feel the same way. Figure 15 displays diagrammatically the distribution in needs across the carers, highlighting that fewer carers feel that they have more than 12 definite needs, with only 7 carers stating that they have 12 or more unmet needs.

When analysing the areas that were reported as a need, 40 carers stated a need for more information, 34 stated a need for more resources and 10 reported a need for counselling. The mean number of resource needs was greater than for information, however, when the number of items within each group is taken into account, the mean value for information needs is greater. This implies that a carer is more likely to report a need for information but when they felt that they had an unmet resource need, they were more likely to report a greater number of resources that were felt to be definitely required.

The need for counselling was reported to be low, with 46 feeling that it was not required and only one carer reporting the maximum number of counselling needs.

This finding suggests that for future interventions, carers are more likely to require more resource and information help in order to meet their unmet needs.

6.2.4 Discussion of the FSS Results

The results of the total number of sources of support felt to be available to the carer are displayed in Figure 16. The histogram, displayed in Figure 16, shows that there is a wide variation in the number of sources of support available to the carers, with the

majority of the carers reporting more than 25 sources of support. The mean number of sources of support from the 57 carers that completed the FSS was 38.09. This implies that as the maximum number of sources of support that could have been reported was 90, the average carer reported that less than half of the maximum number of sources of support was available to them. This seems to suggest that the carers within this population did not have as much support available to them as could be possible. Despite this, there were carers 18 carers that reported 45 or more sources of support available to them, indicating that 32% of carers had more than half the maximum number of sources of support possible to measure using the FSS. This clearly describes the variation of support that carers feel they have had over the previous 3-6 months. Hassall et al found that the higher the FSS total score, the less carer stress was found to be reported using the PSI, displayed with a correlation of $r=-0.485$, $p=0.001$. It would be interesting to assess whether the same relationship will be observed using these results.

The subgroups display the satisfaction that the carers felt towards each area of support. The results shown in Table 19 displays that partner and formal support was reported to have the most satisfaction of how helpful it is to them. Satisfaction of helpfulness was reported to be lowest for the support provided by social organisations and professional services. This is in contrast to the results found by Levy et al, stating that the 132 mothers of children with a disability rated the helpfulness of professional support as being the highest.(Levy et al 1996) This implies that the results identified within the FSS differ from previous findings, with the support provided within a clinical environment not being viewed as helpful as reported by other studies. The reason being may be due to the carers within this study only just having come into regular contact with clinical professionals compared to carers whose child has had a longer standing disability diagnosis. The findings do seem to suggest that not all carers report a lot of support available to them and that the helpfulness provided by professional services could be improved.

6.2.5 Discussion of the Brief COPE Inventory Results

The Brief COPE Inventory was used to examine three different groups of coping strategies. As the questionnaire was designed to assess to how much each carer used the specific coping strategy, a total score could not be generated, as it would be unclear

as to what the score would show. In order to compare the three groups of coping strategies against each other, the mean had to be standardised in order to account for the uneven weighting of each type of coping strategy. Therefore, the mean score was divided by the number of items within each group. This then allowed for an accurate comparison of scores between each subgroup. This was needed, as the Brief COPE Inventory was not able to produce a single score. The standardised mean showed that carers were more likely to use problem-focused coping strategies, producing a score of 4.93. The next most commonly used coping strategy was emotional-focused. The least likely coping strategy to be used by carers was dysfunctional. Cooper et al found that carers were unlikely to use both emotional and dysfunctional coping strategies, and this could explain why there are differences between the two scores; a carer generally uses one or the other. Cooper et al additionally found that dysfunctional coping strategies predicted carer burden.(Cooper, Katona & Livingston 2008) It will therefore be interesting to assess whether dysfunctional coping strategy is associated with increased carer stress scores.

6.2.6 Discussion of the Internal-External Locus of Control Results

Table 21 displays the summary statistics of the results from the Internal-External Locus of control questionnaire. The maximum range of scores for this questionnaire is 0-23. The carer population being studied scored a range of 6-22, implying that higher scores were achieved towards the external locus of control end of the continuum than the internal end. This suggests that carers are more likely to have more external locus of control. However, the overall mean for the study population was 12.33, which lies in the middle of the theoretical continuum created by Rotter. Rotter stated that the mean value will be between 10-12, making this study's results agree with this statement.(Rotter 1975) The mean score along with the median of 12 and mode of 10 suggests that the population is normally distributed across the internal-external continuum. The frequency of the scores can be seen in Figure 18. This displays that the majority of the carers did score within the middle of the continuum, supporting the suggestion that the population was normally distributed. This finding was expected as Rotter stated that this was the typical finding.(Rotter 1975) When the carers were categorised into either internal or external locus of control, the mean value for the internal was towards the top end of the internal range (0-11) whilst the mean external

value was towards the bottom end of the external range (12-23). Perrin et al. found that mothers of children with a chronic physical condition, including those who had a child with a seizure disorder, reported lower scores for internal locus of control and higher scores for external, overall suggesting that the mothers in the population had an external locus of control.(Perrin & Shapiro 1985) These findings are in contrast to this study, as although 55% of the carers were categorised as having an external locus of control when the median value was used as a cut-off, the carers appeared to score more highly in the internal category and lower in the external category. The difference in the group sizes was due to the 5 carers that scored exactly 12. This supports the notion that most of the carers score within the middle of the continuum. Therefore, it was felt that the actual locus of control score will be used for further analysis rather than categorising the carer as having either an internal or an external locus of control as the groups were so even and the extremes of the continuum were not made apparent.

6.2.7 Discussion of the GHQ-28 Results

A total of 57 carers completed the GHQ-28 and the mean for the total score was found to be 7.46. This suggests that on average, every carer felt that they were experiencing 7.46 areas of worse health than normal over the past few weeks. Having said this the most frequently reported score was 0. The range in the total scores, intimates that there is wide variation between carers as to whether or not they feel that their health has been worse over the past few weeks.

Goldberg suggested that a threshold value of 4/5 should be used in order to identify psychiatric disorders.(Goldberg et al. 1997) Using this threshold limit, 30 of the carers would be classified as having a potential psychiatric disorder. This corresponds to 53% of the carer population. As the carer population is a non-psychiatric population, this does seem quite high and warrants future investigation. However, a previous study of 35 parents who have a child with epilepsy, found using the GHQ-28 that 65.7% of parents were above the threshold.(Behrouzian & Neamatpour 2010) Another study examining GHQ-28 scores in 50 caregivers to individuals with epilepsy, found that 55% were above the threshold.(Anjum, Chaudhry & Irfan 2010) This indicates that despite the high percentage of carers identified within this study as being above the threshold, other studies have reported similar rates when caring for those with epilepsy.

All but one of the subgroups achieved the maximum range of scores. The only subgroup that did not was the subgroup representing social dysfunction. The mean score was highest for the anxiety and insomnia subgroup, followed closely by somatic symptoms. This implies that when carers report worse health than normal, it appears that they are more likely to report altered health within these subgroups than the other subgroups that are assessed by the GHQ-28. Depression subgroup had the smallest mean value of 0.78 and the median and modal values were 0. However, the maximum score was achieved within this subgroup. This suggests that there were carers that were suffering with severe depressive symptoms but the majority of carers were not.
This questionnaire does not attempt to assess whether or not the potential diagnosis of epilepsy in their child is the cause to the carer's altered health and therefore, it is quite likely that the carers reporting high depressive scores are likely to be suffering with depression that is unrelated to the child's potential diagnosis.

6.2.8 Discussion of the FACES-IV Results

6.2.8.1 Total Circumplex Ratio

From the 54 carers that completed the questionnaire, three families reported a ratio of less than one. This indicates that according to FACES-IV, three families had unbalanced family functioning and therefore more likely to be an unhealthy system. 22 families reported a ratio between one and two. Olson stated that a ratio of one indicated that there was equal balance versus unbalanced score, therefore it could be assumed that these 22 families were functioning adequately. A ratio above one was stated to indicate that the family functioning was healthy, with the higher scores indicating more balanced than unbalanced scores. As the balanced scores represent good flexibility and family cohesion, this is the most desirable for good family functioning. 29 families fell into this category, with 3 families indicating that the ratio score was above three. Overall, it can be assumed that the families, as reported by the carer, had a good level of functioning, with the mean ratio score of 1.96 demonstrating this. This implies that on average, the family environment could be considered as being a supportive system and should be able to respond effectively to the stress that might be generated to a potential new diagnosis of epilepsy in their child.

6.2.8.2 Family Satisfaction Scale

The mean satisfaction level of the families was found to be 53.76%, falling into the 'moderate' satisfaction category. This implies that on average, carers were reasonably satisfied with the way their family responded to situations and the way that they functioned.

The range in family satisfaction was wide, with carers reporting from 'very low' satisfaction to 'very high'. Satisfaction corresponds to how satisfied the carer is with the family. 10 carers reported very low satisfaction, 8 reported low satisfaction, 10 reported moderate satisfaction, 15 reported high satisfaction and 9 reported very high satisfaction. This shows that the category with the highest frequency of carers was 'High' with similar carer frequencies being reported within the other categories. The 'Low' category was least likely to be reported by a carer.

6.2.8.3 Family Communication Scale

The family communication score is a measure of the quality of communication between family members. The range of scores was again very wide, highlighting the wide variety of carer's perception of their family. The mean score given was 64.17%, falling into the 'High' category. 57% of the carers were found to either categorise the quality of communication within their family as being 'High' or 'Very high'. This shows that the majority of the carers within this population reported very good scores for the quality of family communication. This is important as it implies that issues will be discussed between family members, which will result in a unified family meaning of the event. From the family stress models, it was felt that family meanings played a pivotal role in determining the way that a family responds to stress.

These three scores imply that on the whole, there was reasonable satisfaction with family functioning and this was displayed with an adequate average total ratio. Family communication was generally felt to be strong, implying that the average family environment encouraged good family functioning.

6.2.9 Discussion of the SDQ Results

51 carers completed the Strengths and Difficulties Questionnaire (SDQ), representing the carer's opinion of their child's behaviour and emotion. The total SDQ score represents the difficulties that the child has been said to have. It is formed from the four subgroups: Emotional problems, Conduct problems, Hyperactivity and Peer problems. The range of the total SDQ scores can be seen in Figure 24. The mean total SDQ score was found to be 14.67, which lies in the borderline category. Normal values have been determined from a population of 10298 parents, with the mean for the total SDQ score being 8.4.(Youth In Mind 2011) This shows that there is a large discrepancy between the normal SDQ total score and that found by this present study. This suggests that the children within this population are experiencing more behavioural and emotional problems compared to the normal population. This finding is in keeping with previous findings, with studies identifying that behavioural problems in a child diagnosed with a chronic condition are more prevalent and are a cause of parental stress.(Austin, Risinger & Beckett 1992; Hassall, Rose & McDonald 2005; Rodenburg et al. 2007)

However, the majority of the carers gave a total score of less than 14, the upper normal range cut-off value, with 51% of the children being rated as having normal behaviour and emotional health. Despite this, 39% of the carers rated their child as having high levels of behavioural and emotional difficulties, with the highest score being 32 out of a possible 40. This indicates that there were children within the study population that were felt to have behavioural difficulties by their carers, a finding that was identified within the literature review. The subgroup that scored the highest mean score was hyperactivity, the same finding as identified within the normal population data. The normal population stated that the mean score was 3.5,(Youth In Mind 2011) compared to the mean score of 5.86 found within this present study. It should be noted that none of the mean subgroup scores were within the abnormal range.

The mean impact score was found to be 1.73 with the median value being 1. This shows that the majority of the carers felt that their child's behaviour and emotional difficulties were having a negative impact on their child's life. This shows that there is a need to help the carers with their child's difficulties as it is being felt it may be affecting different areas of their life.

6.2.10 Discussion of the PedsQL 4.0 Results

37 children completed the PedsQL 4.0 with the distribution of their total scores seen in Figure 25. The range of the total score was wide, with some children reporting very low quality of life scores whilst others reported very high levels. This clearly displays the wide range of the studied children's perception of their quality of life. However 65% of the children who completed the questionnaire reported a score above 70. This implies that the majority of the children were experiencing good quality of life.

The mean for the total score was 73.04, lower than the normal mean that was reported to be 83.84. However, the mean found within this population was within the standard deviation of the normal mean. This implies that although the mean for this population being studied was less than that found in a healthy population, it was still within the normal limits. Reduced quality of life has been previously reported when a child is diagnosed with epilepsy,(Devinsky et al. 1999) with Modi stated that reduced quality of life can be experienced even if the child has only had one seizure.(Modi et al. 2009) From the fit demographics, seen in Table 15, 69% of carers reported that their child had had a fit in a public place, and it may be that this has affected the child's quality of life. The findings from the paper by Modi along with this study's results, shows that it must be considered that the fits being experienced are affecting the child's own perspective as to what has become difficult for them. The subgroups display where the child's quality of life has been most affected. The results displayed in Table 26 highlight that schooling has been felt as being a principal cause of reduced quality of life as the mean value of 60.54 is the lowest out of the four subgroups.

6.2.11 Discussion of the SIP Results

Table 27 provides an overview of the self-image (SI) and self-esteem (SE) of all the population who completed the questionnaire. The SE is a reflection of the discrepancy between the two SI scores.

The mean of the positive self-image (SI +ve) was higher than the negative self-image (SI –ve), implying that overall the paediatric population had a positive view of themselves rather than a negative image. However, the range of the negative self-image was wider, showing that there is more variation in the negative image profiles within this population. Patients reported the lowest score for negative self-image, which shows that some do not view themselves negatively at all. The same can be said

conversely with the positive self-image scores, with the highest score being reported, which corresponds to patients having a very strong positive self-image. Neither SI+ve nor SI –ve recorded scores that would correspond to a very poor self-image, which suggests that the population did not perceive themselves in terms of very negative attributes without any positive qualities.

The mean SE score being 8.46 strengthens this claim. This represents that the discrepancy between the two scores was on average, low. A low SE score implies that there is little difference between the two SI scores, suggesting that few patients felt that they would like to be different to how they are now. This suggests that on average, the self-esteem of the population was high. However, the range of self-esteem was wide, with some patients reporting a much wider discrepancy between the two SI scores, suggesting that there were patients recruited with a lower self-esteem than others.

The sense of difference (SD) score reflects how unique that individual feels that they are. Butler states that the higher the SD score, the more separate that individual will view their self, and this has been suggested to be related to lower self-esteem.(Butler 2001) The range of SD scores achieved in this population is the maximum range that could be achieved, highlighting again that there is wide variation in how unique that individual feels that they are. The mean, median and modal values suggest that there is a negative skew for the SD scores, implying that most of the population did report a higher rather than lower SD score. This suggests that the population surveyed did view themselves as being more unique, which may reflect the fact that they are currently being queried with possible epilepsy.

Butler provided normative data for SI +ve , SI –ve and SE scores, based upon the age and gender of the child. He stated that any value that was 2 standard deviation below the normal SI +ve value or 2 standard deviations above the normal SI –ve value or normal SE value would be a cause for concern.(Butler 2001) The data collected from this study population showed that no scores obtained were a cause for concern with all study participants obtaining scores that were within 2 standard deviations from the normal data. The data obtained from this study population along with the normal data as provided by Butler that enabled this comparison to be made, can be seen in Appendix 3.

6.3 Overall Discussion

This study builds upon the literature surrounding carer stress when their child is diagnosed with epilepsy. From reviewing the literature of carer stress and epilepsy, only four studies were found to identify this relationship when the diagnosis of epilepsy in their child is new. To the author's knowledge, this is the first study that examined carer stress when the diagnosis of epilepsy in their child was a potential possibility and so these findings are novel.

The study was carried out to assess whether carers were experiencing stress when their child has a potential new diagnosis of epilepsy as well as investigating the variables that may be associated with increased carer stress. This interim study aimed to recruit carers whose child had not yet been diagnosed with epilepsy, but who had all been referred with this being a suspected possibility. In order to assess carer stress, two measurements were employed, a quantitative questionnaire, PIP, and a semi-structured stress questionnaire. To determine whether carer stress was related to specific variables, the main carer completed 7 different questionnaires. These were the GHQ-28, Brief COPE Inventory, FACES-IV, SDQ, FSS, FNS and the Internal-External locus of control. Each questionnaire was chosen to measure a different variable. In addition to the carer completing questionnaires, the child that had been referred with a potential diagnosis of epilepsy completed questionnaires, although this depended upon their age. The two that could have been completed by the child were the PedsQL and the SIP. The study builds upon a pilot study that was completed in August 2010. The data that has been collected occurred over a 12-month period and will contribute to the main study, due for completion in August 2013.

Previous studies have identified differing levels of parenting stress when their child is diagnosed with epilepsy. Although it may seem intuitive to assume that carers will be feeling stressed when their child has been given a diagnosis of epilepsy, studies that examined parental stress with children who have newly diagnosed epilepsy, reported that the stress levels were low,(Ferro et al. 2010) with only 7% of parents experiencing more stress than a normal population.(Modi 2009) However, other studies have reported higher levels of parenting stress to be present in parents who have a child diagnosed with epilepsy compared to those diagnosed with asthma.(Chiou & Hsieh 2008b)

It was clear that the literature gave conflicting views on whether a child being diagnosed with epilepsy posed a risk to the carer experiencing more stress. Posited reasons for the variation in carer stress between the studies were epilepsy-related factors, mainly seizure frequency and duration of diagnosis.(Austin & Caplan 2007; Modi 2009) This heightened the importance of the present study in collecting information about the fits being experienced by the children. There appeared within this population to be a wide range of fit frequencies, with 39% of carers reporting that their child has only had a maximum of 4 fits whilst 25% reported that the fits occur everyday. This indicates that there was a wide variation in fit frequency; making it an appropriate population to study, as they do not all share a particular characteristic that has been said to relate to carer stress.

However, previous studies have looked beyond epilepsy-related factors and have attempted to examine whether the environment that surrounds the child and carer contributes to the likelihood of a carer experiencing stress. This ideology stems from the family stress theories. It has been identified that a carer with more available resources, specific coping behaviours, better personal health and a cohesive family environment, is related to reduced carer stress. Therefore, it was hypothesised that a carer with more resources, such as more sources of support and fewer unmet needs, a balanced family functioning, fewer personal health problems and the use of coping strategies would result in less carer stress.

The results from this study were obtained from 59 carers, although the number of completed questionnaires differed. The response rate was 45%, with 92% of the carers taking part being female. The homogenous representation of carer gender was not reflected in the demographic analysis, with varying levels of education, different family structures and number of children living at home. This thesis set out to answer three individual aims. Each aim was successful in being addressed and they shall be discussed below.

Aim 1: To explore whether carers are experiencing stress when their child is initially presenting with a potential diagnosis of epilepsy in an outpatient department.

The hypothesis was that carers would be experiencing stress. The results obtained from this study indicated that carers were reporting high stress scores when these were compared to PIP scores obtained in previous studies. The median stress score for PIP-d was 90 and 96 for PIP-f. This implies that carers were scoring highly in both domains, with the health-related events frequency causing more carer stress than their perceived difficulty. However, the range of scores indicated that carers differed as to whether they were experiencing stress. The scores for the stress caused by the difficulty of the illness-related events ranged between 46-174 and the stress caused by the frequency of the events ranged between 56-154. The large range of stress scores was also identified within the semi-structured stress questionnaire, ranging from 0-69. It was additionally found that by using each carer's average PIP score, 43% of the carers were stressed when the categorisation value was set at 90. The percentage of those categorised as being stressed, was higher when using the semi-structured stress score. A cut-off value of 10 was said to indicate stress above the normal population, making 75% of the carers belong to this category.

The median values and the percentage of carers categorised as being stressed, suggests that over half of the carers were generally feeling stressed however the range of scores implies that there is a wide variation in whether a carer is stressed when their child initially presents with a query epilepsy diagnosis.

In order to compare these values to previous findings, the mean values of the PIP-f and PIP-d scores were used due to previous studies presenting the mean scores for each domain. In the present study, the mean scores for PIP-d and PIP-f were 93.11 and 95.54 respectively. It was found that by combining the mean scores from the 7 previous studies, as seen in Table 11, that the average values were PIP-d (88.5) and PIP-f (92.4). Using the combined mean scores indicated that this present carer population were reporting higher levels of stress compared to previous findings.

In summary, it would appear that the carer population were experiencing stress when they were initially presenting in an outpatients department. However, due to neither questionnaire having a clinically set cut-off value to indicate stress, the percentages of those classified as being stressed can only serve as an estimation of the prevalence of carer stress.

Aim 2: To assess if the response to the different variables being measured varies with each family enrolled in the study

The hypothesis was that there would be a range of different questionnaire scores given by the population studied: the carer and their child. All variables were measured by using different questionnaires and it was found that there was a wide range of scores in each questionnaire given by the 59 carers and the 37 children that took part in the study. Summary statistics were generated for each questionnaire to assess the distribution of the total score as well as indicating whether there were differences in the scores of each subgroup for each questionnaire. No statistical analysis was performed on the subgroups to assess for statistical differences in the subgroup scores, or to examine whether there were associations between the subgroups and the carer stress score. It was felt more important to focus upon the overall score as stated by the total score, to indicate the relationships that may exist between factors and carer stress.

It was found that the mean number of unmet needs per carer was 5.95, highlighting that carers could benefit with extra assistance, information being the most reported subgroup of need. This agrees with previous studies that have found that unmet information needs were often reported by carers of children who have been diagnosed with epilepsy.(McNelis et al. 2007; Shore et al. 2009) The FSS showed that the mean number of sources of support was 38.09. Hassall et al demonstrated a correlation between the total FSS score and reduced carer stress,(Hassall, Rose & McDonald 2005) however, although a negative correlation was found in this study, the correlation was insignificant and very weak. This shows that this study did not find any association between an increased number of sources of support and carer stress.

The scores reported on the carer's locus of control were normally distributed across the theoretical continuum, with the mean score being 12.33. There did appear to be more carers categorised as having an external locus of control when the median value (12) was used as a cut-off. However, due to the scores being normally distributed, the difference in groups was due to the 5 carers that reported a score of 12. It was felt that due to the distribution of scores, it would be more useful to correlate the actual locus of control score rather than whether the individual was classified as having an internal or external locus of control. Despite this, there was no association found between locus of control and carer stress.

The Brief COPE Inventory showed that the problem-focused coping strategy was used most often, displayed by the standardised mean of 4.93. This was compared to dysfunctional coping strategies that were reported to be used the least often, displayed by the standardised mean of 3.03. Cooper et al suggested that the two coping strategies do not appear to be used together.(Cooper, Katona & Livingston 2008) This may account for the difference between the scores, however statistical difference was not assessed.

The GHQ displayed that the average carer reported a score of 7.46, with the median value being 6. Both of these are higher than the 4/5 threshold suggested by Goldberg et al as a way to indicate ill mental health.(Goldberg et al. 1997) Using this threshold, 53% of the carers were found to be suffering with ill mental health, this figure agreeing with previous studies.(Anjum, Chaudhry & Irfan 2010; Behrouzian & Neamatpour 2010) There was a wide range of GHQ scores given, with the smallest mean being found in the depression subgroup. This was felt to represent carers that may previously be experiencing depressive symptoms that was unrelated to the child's potential diagnosis. It will be interesting once the main study is complete, to assess whether this trend still occurs as previous studies have found increased carer depression when their child is diagnosed with epilepsy.(Lv et al. 2009; Rodenburg et al. 2005; Shore et al. 2002) A significant correlation was found between the total GHQ score and parental stress and shall be examined later in the discussion.

The total circumplex ratio identified that the average family had good functioning due to the ratio being above 1,(Olson 2011) with the mean value being 1.96. Very few families reported poor family functioning. The health of family communication was found to be higher than the carer's satisfaction of their family functioning, but neither result produced a significant correlation with carer stress. When these three scores were correlated with carer stress scores, insignificant weak correlations were found, suggesting that the family environment was not associated with carer stress. This is in contrast to previous studies that found that poor family functioning was associated with increased carer stress.(Camfield, Breau & Camfield 2001; Duffy 2011; Rodenburg et al. 2007) As only three families were found to have a poorly balanced family function and that the average satisfaction of the family was moderate with the average quality of communication being good, the lack of association may be due to most carers feeling that their family environment is positive.

The SDQ found that carers classified 39% of the children as having abnormal behavioural difficulties. The mean SDQ total score was 14.67, and this is within the borderline range of having more difficulties than would be expected in a healthy population.(Youth In Mind 2011) It has been previously identified that children with epilepsy are reported as having more behavioural difficulties compared to the normal population.(Austin & Caplan 2007; Austin et al. 2004a) This may account for the seemingly high percentages of children with a behavioural difficulty.

The PedsQL identified that the mean quality of life was lower than the normal population, 73.04 compared to 83.84. This value does suggest that the children enrolled in the study were experiencing a lower quality of life than would be expected. The range in the PedsQL score was wide, highlighting that some children felt that their quality of life was good whilst others did not. Schooling was reported as having the lowest mean in the subgroups, suggesting that this was felt by most to be causing a reduced quality of life. Reduced quality of life has been reported in children who have been diagnosed with epilepsy.(Devinsky et al. 1999; Modi et al. 2009) The mean score was still within the normal range although was lower, and does indicate that future support should be offered to children with a potential diagnosis of epilepsy in order to ensure that their quality of life is not affected, regardless of whether or not a diagnosis of epilepsy is confirmed.

Although the PedsQL score seemed to suggest a reduced quality of life, this did not appear to be reflected by the SIP scores. The average SIP scores suggested that most children had a positive self-image, mean of 53.71 compared to 28.74 for negative self-image. The average self-esteem score was 8.46, displaying a small discrepancy between the two self-image scores and therefore implying a high self-esteem. When the individual scores were compared to the normal data,(Butler 2001) no child would have been classified as having a score that was a cause for concern.

Aim 3: To assess if there are any associations between specific variables as reported by the carer and their stress scores

The hypothesis was that there would be some variables associated with carer stress. The results obtained proved that this hypothesis was correct; identifying through the Spearman Rank correlation that four questionnaires produced a significant correlation

($p<0.01$) with the average PIP score, with two of the Brief COPE Inventory subgroups also significantly correlating ($p<0.05$) with the average PIP score. The strongest correlation identified was between the average PIP score and total behavioural difficulties in the child ($r=0.544$, $p<0.01$). However, due to all correlation coefficients being below 0.6, the association between the variables and carer stress were all classified as being weak. This meant that despite the strong significance demonstrated in six of the factors examined (GHQ, SDQ, FNS and all subgroups of the Brief COPE Inventory), the coefficient implied that the correlation could be random, therefore, it was not felt appropriate to analyse the results any further. However, the presence of a significant correlation did warrant further investigation, especially to establish if there was a difference in carer stress scores and those of the six factors identified.

The next statistical test used to prove the hypothesis that certain variable were related to increased carer stress was the Mann Whitney U test. This was achieved by categorising the carers as being stressed or not stressed and to then compare the total scores of each questionnaire. The Mann Whitney U test identified that there was a significant difference, $p<0.05$, between carers categorised as being stressed and not stressed for the total GHQ score, the total SDQ score, total FNS score and the two Brief COPE Inventory subgroups emotional and dysfunctional coping.

A binary logistic regression model found that both the FNS and dysfunctional coping strategy were of equal predictive power, creating two predictive models of carer stress: SDQ + FNS or SDQ + dysfunctional coping strategy. This implies that higher behavioural difficulty scores of their child, along with higher unmet needs or more use of dysfunctional coping strategies, predicts carer stress.

Future work

The binary regression suggested that the completion of the SDQ along with either the FNS or the Brief COPE Inventory could be used instead of the complete battery of questionnaires, as the combination of either two questionnaires was able to predict carer stress. This is important as it implies that in the future, a carer could complete the PIP along with either SDQ + FNS or SDQ + Brief COPE Inventory, and the scores will allow for a quick identification of the carers that are more likely to be stressed. This would then allow those carers to be identified and referred for extra help, such as given by an epilepsy nurse specialist, therefore targeting those that need the help the most

from a limited resource. Obviously this would have to be tested with a designed intervention study in order to assess whether the model suggested by the binary logistic regression was effective at identifying the carers that are stressed. This identifies the future work that could be carried out using the results that have been obtained within this study. Additional future work that could be carried out would be analysing the results collected further; assessing for statistical differences between the subgroups and associations with carer stress. It may be that specific subgroups of the questionnaires provide a more powerful predictive model than suggested from these results. There was qualitative information that was collected through the semi-structured stress questionnaire. Unfortunately, these results could not be analysed nor presented within this thesis, but does provide a rich database for future work. It would be interesting if carers regularly expressed a reason for the stress being experienced and this may help to guide the intervention that is needed to help decrease carer stress. The questionnaires completed by the child were collected, however only a descriptive analysis was performed on the results. The reason being that this thesis has focused upon carer stress and to understand whether the factors, as reported by the carer, were related to their stress score. The questionnaires completed by the children will be used by the main study, with future work needed in order to assess whether a child's score within a questionnaire can affect carer stress, or vice versa.

6.4 Limitations

Despite best efforts, only 54 carers, each with their child, were enrolled into the study, producing a total database of 59 carers in all. Although 59 did allow for some analysis other than descriptive, the sample size was still too small to perform any further statistical analysis than was attempted. It should also be noted that of the 59 datasets, there were 8 that were incomplete, reducing the sample size for specific questionnaires. The reason for the incomplete datasets was due to questionnaires not being returned to the author or carers not attending the next scheduled appointment.

It should also be noted, that due to the questionnaires not being validated for children under the age of 6 years, the number of children that completed the questionnaires was low, with only 37 able to complete the PedsQL and 35 able to complete the SIP.

The recruitment rate for the data collection from September 2010 to June 2011 was 41%. It was noted that 51% were not interested in partaking in the study, of whom, 2

carers specifically stated that they were 'too stressed to talk about it' and 3 carers stated that they did not want to partake when they were not sure as to what the diagnosis was. This indicates that there may be respondent bias as the results do not give a true representation of all carers who have a child with a potential new diagnosis of epilepsy. In order to try and reduce respondent bias, enrolling carers who were experiencing a variety of different emotions, it was emphasised that it was not necessary to know the diagnosis in order to take part in the study. However caution was taken, as it is paramount that the carer or the child is aware that the study is voluntary.

The external validity of the findings may be affected, as mentioned above, by the sample of carers who were willing to be involved in the study but also by the location of the study. All carers were those who had a child who had been referred to Alder Hey Children's Hospital. It is therefore reasonable to suppose that the experience of being referred to a tertiary centre may be seen as a more stressful experience than say presenting to their local general practice. This may make it difficult to generalise the findings of this study to parents who present with their children at other locations.

The main study aims to retrospectively create two groups, depending upon the end diagnosis given to the child. Therefore, for this interim analysis, there was no comparison group, with comparison of results depending upon published normative data or previous study findings. The limitation of this was that it could not be accurately assessed whether the results were different from what would be expected in a normal population, and therefore analysis could only be a descriptive review of the results for the FSS, FNS and Brief COPE.

The questionnaires used for this interim analysis were ones that had previously been chosen before the data collection began in September 2010, following from the pilot study that had been instigated the previous year. It was identified within a review of studies examining carer stress, see appendix 2, that there are many alternative questionnaires that could have been used to assess the same variable. The limitation of the PIP is that it does not have a clinically set cut-off value that classified whether or not the individual was stressed. This does mean that the cut-off score of 90 may either be an under or overestimation for the carer stress. However, the cut-off score was determined from the mean of 7 studies as an attempt to make the categorisation as accurate as possible. It should also be noted that the method chosen in categorising

whether a carer was stressed or not, involved averaging their score from the difficulty and frequency domain. It was found that this method caused 6 carers to be categorised as being stressed and 2 carers as not being stressed, despite all producing only one domain score as being >90. This may mean that there was an over-estimation of the number of carers stressed or that the 2 carers who were not, should have been categorised as being stressed due to one domain score indicating that they were.

It was apparent after reviewing the alternative measures that, it might have been more appropriate to use the Parenting Stress Index (PSI), as Abidin states that a score of 90 signifies clinically significant parenting stress.(Abidin 1992) This would have allowed for a more accurate categorisation of carers who were stressed. Despite this, the PIP did report higher reliability and validity compared to the PSI, see Appendix 1, and the results obtained from the semi-structured stress questionnaire also reported high stress scores, strengthening the argument that the cut-off value used may provide a good estimation of carer stress.

6.5 Conclusion

To conclude this thesis, it was apparent from reviewing the literature that parental stress has been found to be associated with their child being diagnosed with epilepsy. Of the studies reviewed, there were many that have examined parental stress when their child is diagnosed with epilepsy, although there have been remarkably fewer studies that have examined parental stress when their child is newly diagnosed with epilepsy. The review identified that there appears to have been no studies that have examined parental stress when the diagnosis of epilepsy is a potential possibility. This study found that carers who were presenting initially with their child who had a potential diagnosis of epilepsy were reporting high stress scores as evidenced by the PIP and the semi-structured stress questionnaire. Of those, 45% were categorised as being stressed when the PIP was used. Using statistical analysis, the factors that were found to be predictive of carers reported as being stressed were higher levels of behavioural difficulties in their child, with higher numbers of unmet needs or the use of dysfunctional coping strategies. These predictive models suggest that for future interventions, carers that report high stress scores in the PIP along with the questionnaires that measure each of these factors, SDQ, FNS and the Brief COPE

Inventory, could be used to target carers that might need extra help to reduce carer stress.

By using the theory from the different family stress models, increasing the support available to counteract these reported factors, could help to reduce carer stress. The ideal result would be to increase the family's ability in maintaining a healthy level of functioning with the result being that positive quality of life for all family members will be observed. However, this will only be able to be assessed with a future intervention study but the results of this study do suggest that the potential diagnosis of epilepsy is a cause of stress to many carers, warranting further investigation in order to eliminate it.

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Appendix 1: Systematic Search Strategy for Critical Appraisal

Search strategies used for critically appraised papers in Chapter Four.

Medline (Pubmed) was searched in July 2011 using the following search strategy:

1. (("Epilepsy" [MESH] AND "Parents" [MESH} AND "Stress, Psychological" [MESH]))
    a. limits: Humans, English, All child:0-18 years

SCOPUS was searched in July 2011 using the following search strategy:

2. (TITLE-ABS-KEY (epilepsy)) AND (TITLE-ABS-KEY (parent*)) AND (TITLE-ABS-KEY (stress*))
    b. limits: Humans, English

CINAHL and PsychInfo (coverage 1887-current) were searched in July 2011 using the following search strategy:

3. (("Epilepsy" AND "stress" AND "Parent*))
    c. limits: Humans, English

Appendix 2: Articles Reviewed for Outcome Measures

Article	Objective	Sample Size	Outcome Measures	Findings
Transition experience of parents caring of children with epilepsy.[1]	Investigate the health-illness transition from a parent's perspective when a child is diagnosed with epilepsy	10 couples	Open questions regarding the family experience of their child being diagnosed with epilepsy	Psychological distress related to stigma, anxiety and worry regarding the unpredictable pattern of seizures.
Parenting stress and childhood epilepsy.[2]	Evaluate parental stress who has a child with epilepsy.	65 parents	PSI-SF, CDI.	45% of parents reported increased stress. Increased parental stress associated with depression and learning disabilities in the child.
The impact of a new pediatric epilepsy diagnosis on parents.[3]	Compare parenting stress in parent who has a CWE and those without epilepsy.	Epilepsy-30 Control-29	PSI, Family stress scale-seizure version, DPD	No significant differences in parents of children with epilepsy to control.
Boundary ambiguity, coping patterns, depression in mothers caring for children with epilepsy.[4]	Examine boundary ambiguity, coping and depression in mothers who have a child with epilepsy.	316 mothers	Boundary ambiguity scale-for children with illnesses, CHIP, BDI	Boundary ambiguity negatively correlated with coping styles I and II in CHIP.
Coping with Dravet Syndrome.[5]	Investigate how parents cope with Dravet syndrome.	24 parents	Semi-structured interview, ICND.	3 stages of distress: Uncertainty of diagnosis, negative parental relationship, parental social isolation.
Parenting stress in parents of children with epilepsy and asthma.[6]	Identify differences in parenting stress between parents who has a child with epilepsy and those with asthma.	Epilepsy-49 Asthma-54	PSI, Illness Condition Severity coding.	Higher stress in parents who have a child with epilepsy
Parents of children with enduring epilepsy.[7]	To try and predict parenting stress from stressors, resources and coping styles when they have a child with epilepsy.	91 parents	PSI, Functional Status II, CBQ, SDS, Nijmegen Child-rearing Situation Questionnaire, FACES, IPOV, UCL, PACIQ Parental Attitude Research Instrument	Parenting stress was related to child's functional status, parental depression, lower levels of parent-child relationship and difficult child temperament.
Parenting stress of children with intractable epilepsy. [8]	Investigate the prevalence and characteristics of maternal stress in families with intractable epilepsy.	52 mothers	PSI, Scales of Independent Behaviour-revised, CBCL	63% of mothers scored in the clinical range of total stress in the PSI. Correlation of total stress score to total problems on the CBCL ($r=0.5$, $p<0.003$).

Article	Objective	Sample Size	Outcome Measures	Findings
Paternal reactions to a child with epilepsy.[9]	Investigate stress in a father when caring for a child with epilepsy.	210 fathers	PPUS, CHIP, BDI,	Higher stress scores in fathers than mothers.
Correlates of behaviour problems in children with epilepsy.[10]	Identifying variables in predicting behaviour problems in children with epilepsy	127- mothers and CWE	Interviews, CBCL, FILE, FIRM, seizure severity	Family stress and seizure frequency were positively correlated to child behaviour problems.
Adaptive functioning in children with seizures.[11]	Impact of maternal anxiety on child's adaptive functioning	56- mothers	Parental Anxiety about Epilepsy Questionnaire, FILE, CRI, STAI, parental Protectiveness Scale, Parental Problem-Solving Directiveness Questionnaire, Vineland Adaptive Behaviour Scale	Family stress increased anxiety at initial time point.

PSI-SF = parenting stress index short form; CDI = Child Depression Inventory; DPD=Daily phone diary; CHIP= coping health inventory for parents; BDI= Beck Depression Inventory; ICND= Impact of Childhood Neurologic Disability Scale.
PSI-SF = parenting stress index short form; CBQ= Child Behaviour Questionnaire; SDS= self rating depression score; FACES= Family Adaptability and Cohesion Evaluation Score; IPOV= Interactional Problem Solving Questionnaire; UCL= Utrecht Coping Checklist; PACIQ-R= Parent Child Interaction Questionnaire revised; CBCL= Child Behaviour Checklist; PPUS= Parental perception of Uncertainty Scale; CHIP= coping health inventory for parents; BDI= Beck Depression Inventory; FIRM= Family Inventory of Resources for Management; FILE= Family Inventory of Life Events and Changes; CRI= Coping Resources Inventory; STAI= State-Trait Anxiety Inventory for Adults.

Alternative Questionnaires That Could Have Been Used

Variable	Questionnaire	Domains (Items)	Reliability (Cronbach's alpha)	Convergent Validity
Carer Stress	Pediatric Inventory for Parents (PIP)[13]	4 (42): frequency 4 (42): difficulty	Total score: 0.82-0.97 Subscales: 0.69-0.92	Total frequency and difficulty compared to: State Anxiety 0.38-0.62; Parenting Stress Inventory subscales 0.29-0.38; health care usage 0.24-0.47.
	Family Inventory of Life Events (FILE)[14] [15]	9 (71)	Total Score: 0.81	Subscale comparison to FES with no to moderate correlations (0-0.42).
	Parenting Stress Index short form (PSI-SF)[16]	3 (36)	Total score: 0.83 Subscales: 0.80-0.87	Good comparison with parent report data and observational data
	Impact on Family Scale (IOF)[17]	4 (24)	Total score: 0.80-0.93 Subscales: 0.46-0.91	Total score to carer quality of life (-0.54 to -0.70); Daily hassels = 0.35; depression = 0.35; family support (-0.43).
Carer Coping	Brief COPE Scale[18]	3 (28)	Emotional focused 0.72 Problem focused 0.84 Dysfunctional 0.75	Emotional focused coping correlated with number of confidents. Dysfunctional coping predicted more carer burden. Carers were unlikely to use emotional and dysfunctional coping.
	Coping Health Inventory for Parents (CHIP)[19]	3(45)	Total score: 0.85-0.94 Subscales: 0.59-0.91	Family integration with FES cohesiveness = 0.21-0.36 Increased perceived helpfulness of coping strategies associated with lower stress. Higher carer burden with lower coping scores.
	Brief Resilient Coping Scale (BRCS)[20]	1(4)	Total score: 0.64-0.76	High BRCS scores correlated with high levels of positive coping resources and psychological well-being.
Carer Health	General Health Questionnaire (GHQ-28)[21]	4 (28)	Threshold of 5/6 resulted in 79.7% sensitivity, 79.2% specificity and 54.7% positive predictive value.	Compared against the ICD-10 diagnosis generated from CIDI-PC.
	Becks Depression Inventory (BDI)[22]	2 (21)	Total score (0.81-0.86)	BDI total score correlated with clinical rating (0.60-0.72) and Hamilton Psychiatric Rating Scale for Depression (0.73-0.74)
	State-Trait Anxiety Inventory (STAI)[23]	2 (40)	State subscale: (0.91-0.95) Trait subscale: (0.96)	BDI total score with the subgroups 0.65-0.66

Variable	Questionnaire	Domains (Items)	Reliability (Cronbach's alpha)	Convergent Validity
Carer Locus of Control	Internal External Scale[24]	2 (29)	Scale: (0.69-0.73)	Reid and Ware's fatalism subscale (r=0.55) and Levenson's Chance subscale (r=0.45)
	Internal Control Index (ICI)[25]	1 (28)	Total score: (0.84-0.85)	Correlated with Rotter's I-E Scale (r=-0.385, p<0.0001)
	Parental Locus of Control Scale (PLOC)[26 12]	5 (47)	Total score (0.92) Subscales (0.65-0.77)	Did not correlate with Parent Attribution Test or the Parenting sense of competence efficacy scale.
Carer Support	Family Support Scale (FSS)[27]	5 (18)	Total score: (0.77-0.85) Subscales: (0.36-0.76)	Total support compared with: FACES cohesion 0.27; PSI total -0.26.
	Family Stress and Support Inventory (FSSI)[28]		FSSI support (0.78) FSSI stress (0.68)	FSSI stress scale correlated with FILE 0.50. FSSI support scale correlated with FIRM 0.10
	Perceived Social Support (PSS)[29]	2(20)	Total score: (0.88-0.90)	PSS subscales were negatively correlated to Langer scores; -0.27,-0.29.
Carer Needs	Family Needs Survey[30]	3 (35)	Total score: 0.91 Counselling needs:0.87 Resource needs:0.84 Information needs:0.78	Counselling needs correlated with FAM-BF and PSI-SF; 0.30 and 0.27 respectively.
	Family Inventory of Resources for Management (FIRM)[31]	4 (69)	Total instrument: 0.89 Subscales: 0.62-0.85	Family Environmental Scale subscales positively correlated with FIRM subscales.
Family Functioning	Family Adaptability and Cohesion Evaluation Scale –IV (FACES-IV)[32]	6 (42)-FACES 2(20)-Satsifaction and Communication	FACES: Disengaged: 0.79-0.87 Enmeshed: 0.77-0.81 Rigid: 0.65-0.83 Chaotic: 0.76-0.93 Cohesion: 0.89 Flexibility: 0.80 Communication: 0.90-0.94 Satisfaction:0.90-0.94	Disengaged -0.81 with SFI; -0.27 to -0.82 with FAD. Cohesion 0.88 with SFI; 0.40-0.85 with FAD Flexibility 0.68 with SFI; 0.21-0.63 with FAD Chaotic -0.62 with SFI; -0.20 to -0.63 with FAD Satisfaction 0.86 with SFI; 0.84 with FAD.
	Family Environment Scale (FES)[17]	10 (90)	Cohesion:0.76-0.85 Subscales: 0.48-82	Cohesion compared to FAD affective involvement subscale 0.68, FACES cohesion 0.74-0.86
	Family Assessment Device (FAD)[17]	1 (60)	General functioning: 0.82-0.92 Subscales: 0.68-0.84	General functioning compared to: FACES-II adaptability:0.50 FACES-II cohesion:0.61

Variable	Questionnaire	Domains (Items)	Reliability (Cronbach's alpha)	Convergent Validity
Child Behaviour Problems	Strengths and Difficulties Questionnaire (SDQ)[33]	5 (25)	Total score: 0.82 Subscales: 0.57-0.85)	SDQ correlated with Rutter questionnaire (r=0.88)
	Child behaviour Checklist (CBCL)[34]	9 (99)	0.84-0.97	Good validity with the Eyberg Child Behaviour Inventory (r=0.66-0.82).
	Child Behaviour Questionnaire (CBQ)[35]	15 (118)	0.67-0.94	Correlation between mothers and fathers agreement of the CBQ (r=0.28-0.79)
Child Quality of Life	Pediatric quality of life questionnaire (PedsQL 4.0) [36]	4 (23)	Total score=0.88 Subscales=0.68-0.83	Morbidity indicators compared to total score: care needed -0.24; school missed -0.22; work concentration -0.28. Known groups analysis showed healthy children performed better than a chronic health condition 0.73 effect size.
	Child Health Questionnaire (CHQ-CF87)[37]	12 (87)	Subscales: 0.56-0.90	Mean CHQ-CF subscales were lower in subgroups with 1+ reported conditions compared to not conditions.
	KINDL[37]	6 (24)	Total score: 0.84 Subscales: 0.63-0.76	Total KINDL correlated with General wellbeing in the Child Health Questionnaire (0.72); Life satisfaction of the FLZ (0.69); SF-36 emotional wellbeing (0.64).
Child Self Image and Self Esteem	Self Image Profiles (SIP-C, SIP-A)[38]	3 (25)	SIP-C self image (positive & negative)=0.69 SIP-A self image: positive=0.69 negative=0.79	Compared against SPPC. SIP-C (self esteem)=-0.24 SIP-C (+ve image)=0.28 SIP-C (-ve image)=-0.28 SIP-A (self esteem)=-0.28 SIP-A (+ve image)=0.29 SIP-A (-ve image)=0.20
	Self Concept Scale (SCS)[39]	6(60)	Total score = 0.91 Domains = 0.74-0.81	Self-Esteem Inventory (Coppersmith)=0.85 Tennessee Self concept scale=0.51-0.61
	Self Perception profile for Children (SPPC)[39]	5(36)	Global self worth=0.78-0.84 Subscales=0.71-0.86	Not Reported

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Appendix 3: Self Image Profile Results Divided by Age and Gender

SIP-Children Results

Females	**7yrs (N=5)**	Norm	**8yrs (N=1)**	Norm	**9yrs (N=3)**	Norm	10yrs (N=0)	**11yrs (N=1)**	Norm
SI+ve	**61 (6.56)**	55.57 (7.86)	**55**	53.51 (12.46)	**57.33 (13.43)**	57.22 (8.11)	0	**40**	51.64 (9.70)
SI-ve	**17.8 (14.46)**	25.13 (13.92)	**16**	23.35 (13.85)	**22.67 (9.45)**	23.66 (12.49)	0	**32**	19.95 (12.85)
SE	**8.2 (19.78)**	21.21 (13.64)	**30**	21.98 (18.70)	**6.33 (11.37)**	26.00 (17.07)	0	**1**	31.11 (15.41)

Males	**7yrs (N=3)**	Norm	8yrs (N=0)	**9yrs (N=1)**	Norm	**10yrs (N=3)**	Norm	**11yrs (N=1)**	Norm
SI+ve	**45.67 (23.07)**	47.87 (9.77)	0	**45**	51.35 (9.03)	**52.33 (5.77)**	47.78 (11.82)	**57**	53.40 (9.13)
SI-ve	**26.67 (24.44)**	26.31 (13.78)	0	**10**	25.82 (13.73)	**22.67 (14.36)**	26.07 (11.20)	**36**	25.44 (13.07)
SE	**8.67 (2.31)**	29.00 (24.79)	0	**0**	27.33 (20.72)	**4.33 (11.15)**	31.51 (19.57)	**9**	33.45 (20.84)

SIP-Adolescents Results

Females	**12yrs (N=1)**	**Norm**	**13yrs (N=1)**	**Norm**	**14yrs (N=3)**	**Norm**	15yrs (N=0)	**16yrs (N=1)**	**Norm**
SI+ve	**60**	50.04 (7.84)	**42**	46.41 (8.20)	**53.0 (10.82)**	44.18 (9.10)	0	**54**	44.22 (10.45)
SI-ve	**37**	29.20 (12.10)	**55**	34.59 (10.27)	**31.67 (16.2)**	34.85 (11.80)	0	**45**	37.67 (8.25)
SE	**20**	30.54 (12.12)	**16**	38.92 (15.36)	**8.67 (9.71)**	39.81 (15.86)	0	**6**	40.14 (19.13)

Males	**12 yrs (N=3)**	**Norm**	**13yrs (N=2)**	**Norm**	**14yrs (N=3)**	**Norm**	**15yrs (N=3)**	**Norm**	**16yrs (N=0)**
SI+ve	**51.33 (12.01)**	48.33 (11.87)	**61.0 (8.49)**	45.24 (10.36)	**53 (4.58)**	44.89 (9.25)	**54 (17.35)**	45.38 (10.34)	0
SI-ve	**38.67 (17.21)**	28.20 (12.850	**24.5 (17.68)**	29.22 (12.69)	**43 (10.15)**	30.29 (10.21)	**27 (4.36)**	32.16 (10.60)	0
SE	**-3.33 (8.51)**	32.06 (18.30)	**8 (4.24)**	32.30 (14.73)	**-3.33 (5.86)**	36.16 (16.86)	**5 (8.66)**	34.43 (15.48)	0

Appendix 4: Binary Regression Workings

Step 1: Dysfunctional Coping score was added with the total SDQ score

Variables in the Equation

		B	S.E.	Wald	df	Sig.	Exp(B)
Step 1[a]	dysfunctional_cope	.180	.080	5.019	1	.025	1.197
	SDQ	.128	.056	5.282	1	.022	1.137
	Constant	-4.644	1.613	8.286	1	.004	.010

a. Variable(s) entered on step 1: dysfunctional_cope, SDQ.

Result: Both remained significant and therefore were kept in the equation.

Step 2: Dysfunctional Coping score+ total SDQ score + total FNS score

Variables in the Equation

		B	S.E.	Wald	df	Sig.	Exp(B)
Step 1[a]	dysfunctional_cope	.145	.081	3.156	1	.076	1.156
	SDQ	.129	.061	4.443	1	.035	1.138
	FNS	.146	.084	3.048	1	.081	1.157
	Constant	-4.803	1.692	8.062	1	.005	.008

a. Variable(s) entered on step 1: dysfunctional_cope, SDQ, FNS.

Result: Only the SDQ score remained significant. It was felt that the dysfunctional coping and FNS variables were predicting the same variability and therefore two models were created.

Only with FNS

Step 3: Total SDQ score+ total FNS score

Variables in the Equation

		B	S.E.	Wald	df	Sig.	Exp(B)
Step 1[a]	SDQ	.148	.059	6.383	1	.012	1.160
	FNS	.175	.080	4.786	1	.029	1.191
	Constant	-2.638	.983	7.204	1	.007	.071

a. Variable(s) entered on step 1: SDQ, FNS.

Result: Both remained significant and therefore they were kept in the equation

Step 4: Total SDQ score + total FNS score + total GHQ score

Variables in the Equation

		B	S.E.	Wald	df	Sig.	Exp(B)
Step 1[a]	SDQ	.135	.067	4.078	1	.043	1.144
	FNS	.165	.083	3.996	1	.046	1.180
	GHQ	.029	.074	.156	1	.693	1.030
	Constant	-2.590	.980	6.986	1	.008	.075

a. Variable(s) entered on step 1: SDQ, FNS, GHQ.

Result: The SDQ and FNS remained significant but the GHQ was insignificant and therefore this was removed from the equation.

Step 5: Total SDQ score + total FNS score + emotional coping score

Variables in the Equation

		B	S.E.	Wald	df	Sig.	Exp(B)
Step 1[a]	SDQ	.125	.060	4.299	1	.038	1.133
	FNS	.186	.084	4.879	1	.027	1.204
	emotional_cope	.114	.071	2.549	1	.110	1.120
	Constant	-4.617	1.686	7.500	1	.006	.010

a. Variable(s) entered on step 1: SDQ, FNS, emotional_cope.

Result: The SDQ and FNS remained significant but the emotional coping was insignificant and therefore this was removed from the equation.

Only with Dysfunctional coping

Step 6: Dysfunctional coping + total SDQ+ total GHQ

Variables in the Equation

		B	S.E.	Wald	df	Sig.	Exp(B)
Step 1[a]	dysfuntion_cope	.173	.082	4.389	1	.036	1.189
	SDQ	.119	.060	3.863	1	.049	1.126
	GHQ	.023	.066	.125	1	.724	1.023
	Constant	-4.543	1.635	7.721	1	.005	.011

a. Variable(s) entered on step 1: dysfuntion_cope, SDQ, GHQ.

Result: The SDQ and Dysfunctional coping remained significant but the GHQ was insignficant and was therefore removed from the equation

Step 7: Dysfunctional coping + total SDQ + emotional coping

Variables in the Equation

		B	S.E.	Wald	df	Sig.	Exp(B)
Step 1[a]	dysfuntion_cope	.166	.080	4.335	1	.037	1.180
	SDQ	.117	.056	4.388	1	.036	1.124
	emotional_cope	.100	.072	1.906	1	.167	1.105
	Constant	-6.197	2.056	9.082	1	.003	.002

a. Variable(s) entered on step 1: dysfuntion_cope, SDQ, emotional_cope.

Result: The SDQ and Dysfunctional coping remained significant but the emotional cope was insignficant and was therefore removed from the equation.

End result: Either SDQ + FNS or SDQ + dysfunctional coping were predictive of carer stress.