IMPROVING OUTCOMES IN CHILDHOOD EPILEPSY: THE ROLE OF PSYCHOLOGICAL FAMILY FACTORS

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Declaration

This is to certify that the work I am submitting is my own and has not been submitted for another degree, either at University College Cork or elsewhere. All external references and sources are clearly acknowledged and identified within the contents. I have read and understood the regulations of University College Cork concerning plagiarism.

Signed: _____

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Paper (1): Systematic Review

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Abbreviations: CWE, children with epilepsy

Abstract

Objectives: This systematic review 1) charted the relationships between psychological family factors and psychosocial outcomes in childhood epilepsy, 2) identified which factors have acted as risk or protective factors for psychosocial outcomes, and 3) explored whether psychological family factors contributed to psychosocial outcome, over and above those of seizure and epilepsy variables where the relative significance of all such factors were considered.

Methods: A comprehensive search of six electronic databases was conducted. A total of 30 studies (34 articles), met inclusion criteria for review. Psychological family factors included family environment factors (e.g., family functioning, family resources, family stress/demands, resilience, etc.) and parent/caregiver factors (parental mental health, parenting style, parental worry about epilepsy, parent-child relationships etc.). Psychosocial outcomes pertained to emotional and behavioural adjustment, quality of life, social outcomes and adaptive functioning skills. The adapted *Downs and Black Quality Checklist* was applied to included articles.

Results: Psychological family factors were significantly associated with psychosocial outcomes, with 29/30 studies reporting at least a small association between at least one family factor measured and child outcomes. Parent/caregiver factors were most consistently associated with outcomes, with significant moderate associations at both univariate and multivariate level for all studies. 15/21 studies, which assessed the relationship between both family and epilepsy factors, found that psychological family factors were more strongly

associated with outcomes than epilepsy factors. The overall quality of studies was very satisfactory (mean ± SD = 12 ± 2.3).

Significance: Providing front-line clinicians with appropriate training and tools for assessment and brief intervention, could reduce the negative psychosocial impact that some families and children with epilepsy experience.

Keywords

Epilepsy, Family, Caregiver, Paediatric, Psychosocial, Outcomes, Systematic Review

1. Introduction

1.1. Background and Rationale

Epilepsy is one of the most prevalent chronic illnesses in childhood and is the most common childhood neurological disorder. The Prevalence of Epilepsy in Ireland Report [1] states that up to 37,000 people over the age of five in Ireland has a diagnosis of epilepsy, of which more than 10,000 are children and young people. Epilepsy is a complex condition characterised by abnormal electrical impulses in the brain [2]. This 'electrical storm' results in recurrent seizures, which can be difficult to control, with more than a third of children never gaining full seizure freedom [3]

The impact of living with this chronic condition extends well beyond seizure management and adherence to medication. A plethora of research over the recent decades has shown that although many children with epilepsy (CWE) report living satisfactory lives and function well day-to-day [4] a considerable proportion of CWE report difficulties and deficits in a range of psychosocial domains. These include poorer quality of life, emotional and behavioural functioning, and social competence [5–8]. It was concluded by meta-analysis [9] that CWE are at increased risk for psychopathology, including internalising and externalising emotional and behaviour difficulties in comparison to healthy controls. Another review [10] supported this view, reporting that CWE display more symptoms of depression and anxiety than the general population. Moreover, it has also been highlighted that CWE report poorer outcomes than children with other chronic illnesses [11]. This is in line with the theory that children with diseases of the neurological system are at increased risk for poorer

outcomes than those with more generic diseases [12]. These psychosocial difficulties are reportedly more pronounced in children with a higher rate of seizure frequency [13–15].

As a result of the difficulties outlined, caring for a child with epilepsy can be a stressful time for the entire family. Children with epilepsy share the same family environment and therefore share with them the challenges and difficulties associated with the condition. Epilepsy shares many features with other chronic illnesses, such as daily medication management, frequent medical appointments and limitations to daily activities. However, it has been found to be a more stressful condition for the families affected due to its sporadic and unpredictable nature, which is not found as such in other chronic conditions. Research into the impact of caring for, and living with, a child with epilepsy has shown that a proportion of parents and siblings of CWE report poor psychosocial outcomes and more disruptive family environments than families of children with other chronic illnesses [9]. This suggests that it is both features specific to the epilepsy itself, as well as the chronicity of the disease, that impacts families.

In further support of this, a recent systematic review of quality of life in parents of CWE [16] concluded that these parents experience poorer quality of life, particularly in relation to their mental health, when compared to parents of healthy controls. They further reported that factors relating to the family environment, such as family relationships and financial status, were more robust predictors of quality of life than child and/or epilepsy characteristics. Another recent review [17] reported that more than 50% of parents of CWE report

symptoms of anxiety in clinical ranges, suggesting that these parents are a significantly at-risk cohort.

Although it is undoubtedly evident from the literature that many families struggle as a result of caring for a child with epilepsy, recent research looking at the impact of epilepsy on siblings has produced more positive findings. A recent paper [18] reported siblings of CWE to be considerably resilient and not unduly affected by the disorder, rather that they felt proud and protective of their sibling with epilepsy. These findings were concluded from parental reports, however, are supported by previous research [19] which also included sibling self-reports. These findings may also be related to the characteristics of the specific families themselves in how they, as families, tend to cope with adversity.

In line with this, traditionally, it was thought that child and family outcomes were mediated by seizure frequency/illness factors alone. However, in recent years it has become increasingly acknowledged in the literature that psychological family factors (such as family environment and functioning) play a substantial role in the development and maintenance of child outcomes in epilepsy [12,20]. This is not surprising given similar findings from research with other chronic illnesses [21–23]. Yet, what is particularly interesting is that in some cases psychological family factors have been found to be more strongly associated with such outcomes than seizure variables [4,24].

Still, the evidence to date remains somewhat mixed. Results from an investigation into trajectories of quality of life in childhood epilepsy, assessing

a model of family and psychosocial factors, suggested that family functioning and parental worries were not significantly predictive of quality of life in childhood epilepsy, rather the number of anti-epileptic medications and the presence of emotional and behavioural difficulties in the children themselves were the most consistent predictors [8]. A similar study [4] again looked at trajectories of quality of life in childhood epilepsy, however contrasted the aforementioned findings by underscoring the importance of psychosocial factors, such as parental depressed mood, over seizure and anti-epileptic medication factors in predicting quality of life. Moreover, another study [25] which looked at family functioning but in relation to child emotional and behavioural adjustment also found significant relationships between the variables, over and above seizure variables (frequency, and age of onset), second only to anti-epileptic medications.

Although mixed, these findings potentially serve to act as further stressors for families who are already struggling to adjust to, and live with, epilepsy. More specifically, this means that families, who are negatively impacted by the nature of the disease, are themselves a risk factor for more negative outcomes in their child with epilepsy. Additionally however, it also may mean that families who portray more positive adaptation to epilepsy could promote better outcomes for their child [26]. That said, there is a significant need for the systematic synthetisation of the research, as to date findings have accrued in a somewhat ad-hoc manner. Thus, it is currently difficult to generalise and apply findings, in order to form accurate and reliable conclusions for clinical implications. Establishing such associations could serve to clarify the focus of future child

and family interventions aimed at improving psychosocial outcomes for children and young people with epilepsy. Moreover, such a model of risk and protective factors may act as markers for clinicians involved in epilepsy diagnosis in identifying families for early intervention as a preventative measure.

1.2 Review Aims

To date, and to our knowledge, there have been no reviews evaluating a range of psychological family factors and child psychosocial outcomes in the field of epilepsy. Accordingly, the current review first aims to synthesise the relationship between psychological family factors and psychosocial outcomes for CWE. In doing this, it secondly aims to establish what psychological family factors may act as risk or protective factors for psychosocial outcomes. A third aim of the current review is to establish which psychological family factors contribute to psychosocial outcome, over and above that of seizure and epilepsy variables. Lastly, implications for clinical practice will be considered.

2. Methodology

This review followed the methodological approach recommended for systematic reviews outlined by the *Preferred Reporting Items for Systematic Reviews and Meta-Analyses* (PRISMA) [27]. This included careful preparation and planning of the nature of the review, explicit research questions, a thorough literature search, data extraction and evaluation by two reviewers, synthesis of findings and summation of implications.

2.1 Definition of terms

2.1.1 Psychological family factors

Family factors incorporated for inclusion in the current review include psychological family factors related to the three main domains of factors which were identified through a scoping review of available literature. These include factors related to the family environment (e.g., family functioning, family relationships, family resources, family stress/demands, etc.), family resilience (a family's ability to cope and 'bounce back' in the face of adversity), parental mental health (e.g. depression, anxiety, etc.), and more general parenting (e.g. overprotective/controlling parenting style, parental worry, etc.). There are potentially other family factors which could also be included for review, for example attachment styles, however, as these were not identified through the scoping review, there was not enough information to proceed further with these.

Structural family factors (socioeconomic status, parental employment or education, family composition, etc.) were not included in the current review so as to focus on those psychological factors that are perhaps more amenable to change.

2.1.2 Psychosocial outcomes

For the purpose of the current review psychosocial outcomes include psychological outcomes, such as mental health (e.g. anxiety, depression, behavioural difficulties, self-esteem, etc.), as well as quality of life, social outcomes (e.g. social skills and difficulties), and adaptive functioning skills (e.g. communication, level of independence, and daily living skills).

2.2 Search strategy and study selection

A comprehensive search of Academic Search Complete, MEDLINE, CINAHL, PsychArticles, Psychology and Behavioural Sciences Collection and PsychINFO was completed on September 29th, 2018 using a variety of search terms. These included terms relating to 1) family factors 2) children with epilepsy and 3) psychosocial outcomes. Specifically, the following terms were searched using study title or abstract: "psychological" or "psychosocial" or "emotional" or "mental health" or "adjustment" or "behaviour*" or "behavior*" or "well-being" or "quality of life" AND "child*" or "adolescen*" "child*" or "adolescen*" AND Epilepsy AND "family" or "family factor*" or "family characteristic*" or "family influenc*" or “family functioning.”

The search was restricted to studies published in academic journals and journals that were published in English. No other restrictions were applied. All studies identified for possible inclusion through title or abstract searching were read in full by the main reviewer (EH).

Articles were included if they met the following criteria:

- Published in a peer-reviewed journal.
- Outcomes were focused primarily on children/adolescents (0-18 years).
- Included youth who have an epilepsy diagnosis (as defined by individual study criteria).
- Evaluated the relationship between psychological family factors (identified as the main independent variable) and psychosocial outcome in childhood epilepsy.

Psychological family factors included family functioning, family resilience, parental mental health, and parenting style, while child psychosocial outcomes included mental health, quality of life, social outcomes, and adaptive functioning skills, as outlined in section 2.1.1.

- Used established measures of outcome (e.g., documented reliability/validity, standardized questionnaires).
- Included interpretable data (e.g. results compared to reference group norms or control groups).
- Published in English

Articles were excluded based on the following criteria:

- Focused on participants outside of the specified age range (0-18 years), i.e. studies examining outcomes in adults with epilepsy.
- Focused solely on structural family factors (e.g. socioeconomic status, employment, education, family composition).
- Followed a qualitative design.
- Review articles and case studies were excluded.
- For a paper to be excluded, both reviewers needed to agree that it did not meet inclusion criteria after their independent screenings.

In order to ensure that no relevant articles were overlooked, the reference lists of articles chosen for full-text review were hand searched for additional articles. Following compilation of an initial list of articles deemed relevant for inclusion, articles were discussed with an independent reviewer (CMcC) to reach agreement regarding appropriateness for inclusion.

2.3. Quality check

The scientific merit of all articles identified for inclusion was completed using a modified version of the Downs and Black Quality Index [28]. This process was completed independently by two reviewers (EH and CMcC) and results were discussed, and ratings were finalized. This version of the Downs and Black Quality Index rates papers out of a total of 15 across four subscales, including Reporting (7; e.g. *'Are the characteristics of the patients included in the study clearly described?'*), External Validity (3; e.g. *'Were the patients asked to participate in the study representative of the entire population from which they were recruited?'*), Internal Validity (4; e.g. '*Were the statistical tests used to assess the main outcomes appropriate?'*), and Power (1; e.g. *'Did the study provide a sample size or power calculation to detect important effects where the probability value for a difference being due to chance is less than 0.05?'*). A total of one mark is given for a 'yes' answer, while zero marks are given for 'no' and 'unclear' answers. There are no thresholds or cut-off points for better/poorer quality papers cited by the authors or in corresponding literature, rather, higher scores are indicative of higher study quality. The original index comprised of 27 items; however, the modified version uses a reduced number of items after removing items specifically relating to intervention studies, including blinding, randomization, withdrawals, drop outs and integrity of intervention.

This version has been used in a number of recent systematic reviews and meta-analyses in the current field [4,16,29,30] and can be found in Appendix 2.

2.4 Data extraction and synthesis

Relevant data pertaining to child and family characteristics (participant numbers, mean age, gender, epilepsy characteristics, etc.) were extracted from selected studies. These were formally extracted by one reviewer (EH) and discussed with a second reviewer (CMcC).

Due to significant variability in sample characteristics, measures employed, and available data, a meta-analysis was not deemed appropriate for the current review. Instead, a narrative synthesis of the findings is reported, supported by the presentation of results of univariable and multivariable analysis and level of effect sizes detailed where possible.

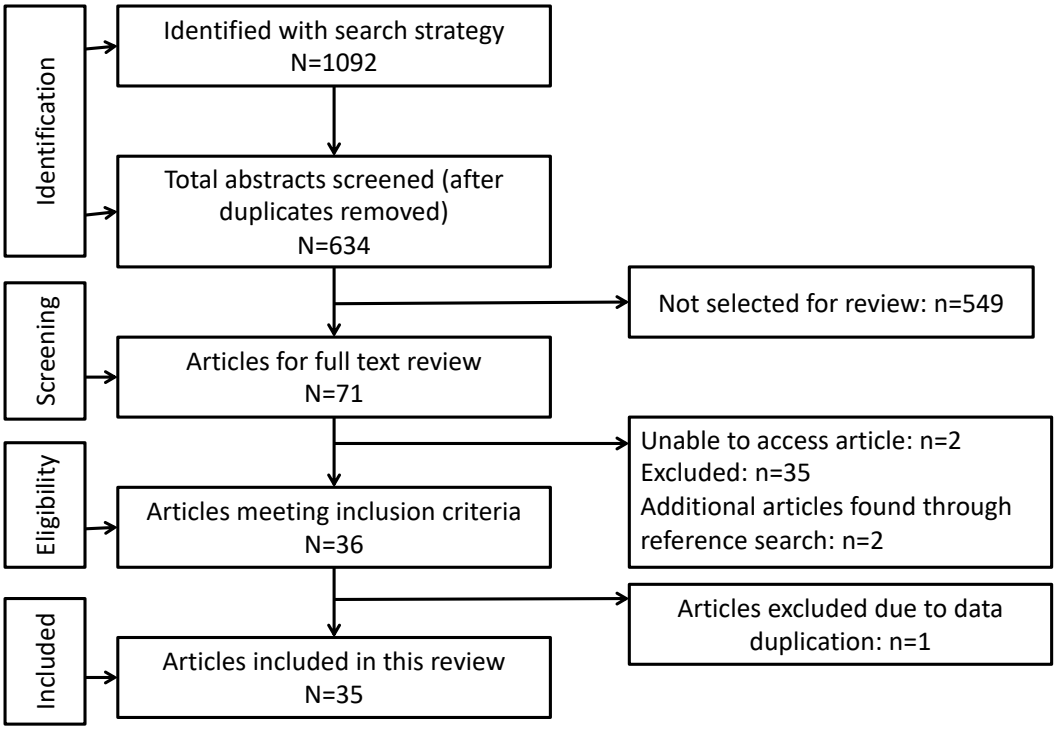
3. Results

3.1 Search results

The search strategy employed returned a total of 634 articles for review of which 71 were selected for full text screening. It was not possible to retrieve the full text for two articles selected for full text review [31,32]. The corresponding authors were contacted in these two cases; however, no response was returned. Of those remaining 69 articles, 35 were excluded based on the inclusion and exclusion criteria outlined. The reference lists of the remaining 34 articles were hand searched by the author (EH) to check for any additional articles that may have been missed during the initial search. Two further articles were identified using this method; meaning 36 articles were included in the current review.

Several of the articles identified for inclusion reported different aspects of the same study and utilised the same participant sample [32–36]. In an effort to avoid data duplication and include the most useful information as possible, the results from these articles were considered as one study – the Health-Related Quality of Life in Children with Epilepsy Study (HERQULES). Two further studies [4,14] reported results from the same sample. The former study [4] reported the combined longitudinal data of both studies, results from this paper were included in the current report. Thus, the final number of studies of which results were analysed for the current review was 30, which included 35 articles. The overall search results and study selection process is presented visually in Figure 1.

Figure 1. *Flowchart of study selection*



3.2 Characteristics of studies included

Table one provides an overview of the characteristics of the included studies, including country of origin, descriptive characteristics of CWE and respondents, design, measures of interest and summary of main findings. The results of univariate and multivariate analysis, as well as level of effect sizes, are also detailed where available.

The studies included were published between 1988 and 2018 and stemmed from research-conducted worldwide. While the majority of studies (n = 18) were carried out in either the U.S.A or Canada, two studies were conducted in Portugal, three in the U.K, two in India, one in the Netherlands, one in Turkey, one in Australia, one in Singapore, one in China and one in Korea. The majority of studies were cross sectional in design (n = 22), with the remainder (n = 8) prospective and one study that reported on secondary data from a randomised controlled trial. Table one also illustrates details pertaining to the characteristics of participants and respondents (parents/caregivers, teachers and children), with an emphasis on child epilepsy details. Seventeen studies collected data from parents and/or caregivers only, six included teacher reports, nine included parent and child self-reports, and one study utilised child self-reports only. Children and adolescents ranged in age from 2 – 18 years of age across 30 studies, with the age range for children remaining unclear in one study [37]. Eight studies concentrated on children with new onset epilepsy. All studies gathered data from children and young people with mixed epilepsy diagnoses and seizure frequencies.

With reference to the assessment of quality of studies (higher scores indicated a higher study quality. A more detailed summary of which can be found in Appendix 3), the overall quality of studies was very satisfactory (mean ± SD = 12 ± 2.3). Reporting quality was found to be quite good (mean ± SD = 6 ± 1.3), as was external validity (mean ± SD = 2.3 ± .8) and internal validity (mean ± SD = 3.7 ± .6). Lastly, one out of 30 studies reported a power calculation for sample size (mean ± SD = .03 ± .2).

Table 1: Summary of studies reviewed

First author (year) Country of origin	Sample size Child % male, mean age ± SD (Respondent)	Design	Standardised family measures of interest	Standardised child measures of interest	Summary of relevant main findings	Quality Index Rating (total scale)
Austin (1988) [38] USA	*n* = 54 43%, 10.7 ± (SD n/a) (parent/caregiver, teacher, child)	CS	Family APGAR, FIRM	CSCS, CBCL	Families with lower family functioning and lower family resources (esteem and communication, mastery and health and social support) had children with more emotional and behavioural difficulties than families of children who did not report these difficulties.	11
Austin (2004) [39] USA	*n* = 224 48%, 8.4 ± 3.0 (parent/caregiver)	PR	FIRM, Family APGAR, PRPC	CBCL	Family mastery negatively predicted total behaviour problems in children with new-onset epilepsy, with results remaining consistent after a 24-month period (baseline: β = -6.27, p < .0001; follow up: β = -4.47, p = .0013). Parent worry (β = 2.97, p = .02) and a greater need for information and support (β = 2.70, p = .03) also predicted behaviour problems at 24 months. However, the relationship between satisfaction with family relationships and behaviour problems did not reach statistical significance at either timepoint.	12
Austin (1992) [40] USA	*n* = 127 50%, 10.5 ± (SD n/a) (parent/caregiver, child)	PR	FILE, FIRM	CBCL	Family stress, mastery and social support were all significantly correlated with child behavioural difficulties at univariate level (p< .001). These associations were also significant at multivariate level (p < .05) with lower levels of family social support emerging as the strongest family predictor of behavioural outcomes (β = -.83, p = .02).	11
Baum (2007) [41] USA	*n* = 109 60%, 12.4 ± 3.3 (parent/caregiver)	CS	FIRM	CBCL	In families with low mastery, children with more difficult temperaments had more behaviour difficulties (β = .29, p = .03). In families with high mastery there was not a significant association between temperament and behaviour difficulties (β = -.11, p = .26).	13
Carson (2016) [42] USA	*n* = 93 49%, 12.3 ± 2.9 (parent/caregiver, teacher)	CS	PAE	CBCL, SBS (teacher rated)	Parental anxiety about epilepsy revealed small to medium correlations with social skills in CWE as reported by parents (r = -.298, p < .01) and teachers (r = -.347, p < .01), and was also moderately associated with parent rated social problems (r = .335, p < .001) but not teacher rated social problems.	10
Chapieski (2005) [43] USA	*n* = 56 64%, 8.6 ± (SD n/a) (parent/caregiver)	CS	PAE, FILE, CRI, PPS, PPSDQ	Vineland-II	Higher levels of maternal anxiety about epilepsy and family stress were related to lower levels of child adaptive functioning in the communication domain (R^2 = 0.11, p < .02, R^2 = 0.08, p < .03, respectively. High levels of maternal anxiety about epilepsy were also associated with the daily living skills (R^2 = 0.26, p < .0001) and socialisation (R^2 = 0.25 p < .0001) domains. Results remained significant for the communication and socialisation domains while parental coping was associated with daily living skills. Parental protectiveness was not associated with adaptive functioning.	13

Chew (2017) [44] Singapore	*n* = 152 52%, 15 ± 1.3 (child)	CS	FRAS	RSS, PSS[a],	Higher levels of family resilience predicted higher levels of self-esteem in a multiple regression (β not stated) for all sample. Multiple mediation analysis showed that adolescents with more severe epilepsy reported lower family resilience than those with low/moderate epilepsy.	14
Conway (2016) [45] Canada	*n* = 115 57%, 11.8 ± 3.8 (parent/caregiver)	PR	QIDS, GAD, Family APGAR, FIRM, FILE	QOLCE	At univariable level, child quality of life scores were associated with caregiver depression (β = -1.75, p < .001), anxiety (β = -1.36, p < .001), higher satisfaction with family relationships (β = 2.21, p < .001), greater resources to aid adaptation to stressful events (β = .77, p < .001) and lower family demands (β = -1.15, p < .001). Fewer resources was the only family variable to significantly predict reduced child quality of life at multivariable level. Although epilepsy variables were most strongly correlated with outcome at univariable level, they were not significant contributors at multivariable level.	13
Ekinci (2016) [46] Turkey	*n* = 53 55%, 11.8 ± 2.6 (parent/caregiver, teacher, child)	CS	FAD, BDI, STAI	CSCS	Family functioning, specifically communication levels (β = -.430, p = .002), affective responsiveness (β = -.389, p = .004), problem solving (β = -.300, p = .03), and roles (β = -.294, p = .04), and maternal depressive symptoms (β = -.392, p = .004) negatively predicted self-concept in CWE. Maternal anxiety was not associated with child self-concept, nor were epilepsy variables.	15
Ferro (2017) [4] Canada	*n* = 505, 486 51%, 11.4 ± 2.1 (parent/caregiver, child)	PR	BCFPI, SSSC	CHEQOL	Fewer symptoms of parental depression, and greater peer and family social support, were identified as the most robust predictors of higher health related quality of life over a 28-month period (all p < .001).	14
HERQULES [32–36]. Canada	*n* = 374 52%, 7.5 ± 2.3 (parent/caregiver)	PR	CES-D, Family APGAR, FILE, FIRM	QOLCE	Parental depression, family functioning, family stress and family resources were associated with poor quality of life two years post epilepsy diagnosis. One study highlighted that family resources moderated the impact of parental depression on child quality of life, while family functioning and family stress mediated this impact [32]. In another study [34], the effect of parental depressive symptoms was mediated through family functioning and family stress, while family resources acted as a moderator between severity of epilepsy and emotional well-being – with children with more severe epilepsy benefitting from increased family resources.	14 [32] 13 [33] 13 [34] 14 [35] 14 [36]
Han (2016) [47] Korea	*n* = 297 61%, 15.4 ± .9 (parent/caregiver, child)	CS	BDI, PSOC, Family APGAR, MSI-R, CRPBI	CBCL-YSR	The strongest factor contributing to internalising problems was high levels of parental depressive mood (β = .233, p < .000) while parental overcontrol (β = .190, p = .001) contributed most strongly to externalising problems. Family functioning, marital stress and conflict over child-rearing were not found contribute any unique variance to adolescent psychopathology, however demonstrated small associations at univariate level.	12
Hoare (1991) [48]	*n* = 108 55%, 10.4 ± 3.0	CS	GHQ, EPASE	PTRBS	Parental mental health status was significantly associated with child behavioural problems as rated by parents (t-test value n/a, p < .01) but not teachers. Parental	7

UK	(parent/caregiver, teacher, child)				attitudes about epilepsy were also associated with child behavioural problems as rated by both parents and teachers (X^2 value n/a, p values ranged from $p < .001 – p < .05$).	
Hodes (1999) [49] UK	*n* = 22 59%, 11.9 ± 2.9 (parent/caregiver, teacher)	CS	CFI, GHQ, SSSI	PTRBS, HSPQ	A moderate, positive, correlation was identified between maternal mental health status and child behavioural difficulties (r = .655, p = .001). Total behaviour difficulties were not associated with maternal emotional over-involvement, level of emotional warmth or number of positive remarks from mothers. However, such difficulties were associated with high levels of criticism (r = .536, p = .006).	14
Kerne (2015) [50] USA	*n* = 97 51%, 12.2 ± 2.99 (parent/caregiver, child)	CS	PAE	Vineland-II	Parental anxiety about epilepsy was significantly negatively associated with both daily living skills (r = -.30, p < .001) and socialisation (r = -.26, p < .01) scores of adaptive functioning at univariate level. Results of a stepwise regression suggested that parental fears about epilepsy also had a direct effect on child adaptive functioning, as well as suggesting that seizures that secondarily generalised affected child adaptive functioning by increasing parental anxiety about epilepsy.	9
Li (2008) [51] China	*n* = 340 60%, 9.1 ± 3.3 (parent/caregiver)	CS	HADS	QOLCE	Parental anxiety was found to be significantly correlated with quality of life in CWE (r = n/a, p < .000), with the more severe the parents' level of anxiety, the lower the level of child quality of life.	7
Malhi (2007) [52] India	*n* = 75 65%, 11.1 ± 1.6 (parent/caregiver)	CS	FES	CSCS, PAAS, CPMS	Control in the family explained 10% of the variance in total self-concept (F = 7.72, p < .007), with higher control associated with higher self-concept. Family cohesion levels [along with the age of the child] explained 17% of the variance of child adjustment (F = 7.67, p < .001), with higher cohesion lending to better adjustment in the child. Lastly, more conflict in the family was associated with higher psychopathology scores, explaining 24% of the variance [along with maternal education], F = 10.94, p < .000).	11
McCusker (2002) [25] UK	*n* = 48 65%, 7.1 ± 2.6 (parent/caregiver)	CS	FRI	CBCL	Higher levels of family conflict were associated with attention/hyperactivity (r = .31, p < .05), social difficulties (r = .3, p < .05) and recreational activates (r = -.3, p < .05). Lower levels of family cohesion were related to attention/hyperactivity (r = -.48, p < .001), social difficulties (r = -.51, p < .001), thought disturbance (r = -.3, p < .05) and school functioning (r = .34, p < .05).	12
McLaughlin (2018) [53] USA	*n* = 108 47%, 11.3 ± 3.7 (parent/caregiver)	CS	ICQ-P	QOLCE	Parental coping (acceptance and helplessness) was significantly related to health-related quality of life in young people with epilepsy at the bivariate level. However, at multivariate level, higher levels of helplessness cognitions in parents predicted lower health related quality of life (β = -.50, p < .01), while parental acceptance of their child's epilepsy was not predictive of health-related quality of life.	11
Mendes (2017) [20] Portugal	*n* = 192 50%, 11.9 ± 3.1 (parent/caregiver, child)	CS	FES, PSS[b], Eurohis-QoL	DCGM	At univariate level, family cohesion showed moderate negative associations with child quality of life as reported by children (r = .39, p < .01) and parents (r = .59, p < .01). Perceived stigma was also produced associations with quality of life as rated by children (r = -.60, p < .01) and parents (r = -.18, p < .05). At multivariate level, family cohesion was positively linked to children's quality of life indirectly by way of negative links with parental perceived stigma. Furthermore, when family cohesion	14

					levels were higher, children and parents reported lower levels of stigma and higher levels of quality of life.	
Moreira (2013) [54] Portugal	*n* = 68 52%, 12.6 ± 2.9 (parent/caregiver, child)	CS	EUROHIS-QOL-8	KIDSCREEN-10	Parental quality of life was not associated with child quality of life at multivariate level (β = -.01, p > .05), explaining 1.5% of the variance of the entire model (Δ R^2 = .015, *F* (total model) = 11.68, p < .05) No information was provided regarding size of association at univariate level.	12
Pal (2005) [55] India	*n* = 64 57%, n/a – range = 6-18 years (parent/caregiver)	RCT	DFSS	CPRS	Parental satisfaction with social support was positively and independently correlated with child behavioural problems (β = .31, p = .03) in a multiple regression, representing the only variable to reach significance. Other variables included a number of epilepsy specific variables.	6
Puka (2017) [56] Canada	*n* = 287 48%, 9.6 ± 2.6 (parent/caregiver)	CS	Family APGAR, FILE, FIRM, GAD, QIDS-SR16	GAD-7, QIDS-SR16, RCADS	Univariable regressions predicting depression in children identified parental depression (β = .462, p = 002 – only factor to remain significant at multivariable level), poorer family relationships (β = .383, p = .01) and poorer family mastery (β = .337, p = .025) as predictors. No predictors of child anxiety were identified. Univariable regressions predicting depression in adolescents identified parental anxiety (β = .321, p = .01 – remained significant at multivariable), poorer family mastery (β = .334, p = .01) and higher family demands (β = .326, p = .01). Similarly, adolescent anxiety was predicted by parental depression (β = .246, p = .05) and anxiety (β = .392, p = .001 – remained significant at multivariable), and poorer family mastery (β = .247, p = .05). No epilepsy factors remained significant for either age group at multivariable level.	14
Ramsey (2016) [8] USA	*n* = 94 59%, 8.4 ± 2.4 (parent/caregiver)	PR	PRPC, FAD	QOLCE, BASC-2	Family functioning predicted social interaction subscale of the QOLCE (X^2 = 8.54, p = .004) but did not predict any other domain of quality of life or emotional/behavioural adjustment. Parental worry was not predictive of any subscale of the QOLCE or emotional/behavioural adjustment. Results analysed after a 25-month period.	14
Rodenburg (2006) [9] Netherlands	*n* = 91 58%, 8.5 ± 2.4 (parent/caregiver)	CS	PSI, PACIQ-R, SDS, FACES, IPOV	CBCL	Parental rejection was correlated with internalising (r = .41, p < .001) and externalising (r = .59, p < .001) problems. Positive parent-child relationship was correlated with internalising (r = -.35, p < .001) and externalising (r = -.64, p < .001) problems. Parental confidence in parenting was associated with externalising problems only (r = -.23, p < .05). Parental depression was associated with internalising (r = .33, p < .01) and externalising (r = .32, p < .01). Family adaptation problems were correlated with internalising (R = .24, P < .05) and externalising (r = .28, p < .05) problems. Marital satisfaction was correlated with externalising problems only (r = -.31, p < .01). Parent-child relationship factors accounted for most variance in all psychopathology, over epilepsy factors.	14
Thornton (2008) [57] USA	*n* = 82 40%, 12.2 ± 3.3 (parent/caregiver)	CS	FAM III	CBCL	Moderate correlations were found between overall levels of family functioning and child total competence (r = -.329, p < .004) and emotional (r = .379, P < .001) and behavioural (r = .478, p < .001) adjustment in CWE.	13

Tse (2007) [58] USA	*n* = 101 52%, 11.2 ± 3.8 (parent/caregiver)	CS	FAM-III	SSRS	Ratings of family functioning revealed a moderate negative correlation with child social skills (r = -.44, p < .001) at univariate level, along with neurological factors. However, at multivariate level abnormal family functioning [and presence of learning disability] were the only factors to predict social skills impairment (OR = 7.83).	14
Williams (2003) [59] USA	*n* = 200 51%, 10.3 ± (SD n/a) (parent/caregiver)	CS	STAI	ICIS	Parental anxiety was found to be significantly, moderately, correlated with decreased quality of life (r = .48, p < .01), second only in strength to severity of comorbid conditions. Parental anxiety was also found to predict quality of life at multivariate level in a stepwise regression model (R^2 =.50, t = 6.8, p < .01).	10
Wu (2014) [60] USA	*n* = 124 60%, 7.2 ± 2.9 (parent/caregiver)	PR	FFS-Seizure, PSI, PRPC, PSS[b]	PEDS-QL, QOLCE	Parent and family stress, perceived stigma, and parent fears and concerns about epilepsy consistently negatively impacted child health related quality of life in a 25-month period and explained an additional 19% of the overall model when demographic and seizure variables were controlled for. In addition, parent stress (β = -.36, p < .05), and fears and concerns (β = -.60, p < .001) contributed unique significance to health-related quality of life in the final model.	14
Xu (2017) [37] Australia	*n* = 77 n/a %, 8 ± 4 (parent/caregiver)	PR	Family APGAR	SDQ	Lower family functioning was associated with psychological distress in children with newly diagnosed epilepsy (OR 1.80 per 1-point decrease on the Family APGAR, mean 14 ± 2, p = .03)	13

n/a = not available, CR = Cross-sectional, PR = Prospective, RCT = Randomised Controlled Trial, FIRM = Family Inventory for Resource Management, Family APGAR = Family Adaptability, Partnership Growth, Affective and Resolve Scale, PAE = Parental Anxiety about Epilepsy questionnaire, STAI = State Trait Anxiety Inventory, FILE = Family Inventory of Life Events and Changes, CRI = Coping Resources Inventory, PPS = Parental Protectiveness Scale, PPSDQ = Parental Problem-Solving Directedness Questionnaire, Family Resilience Assessment Scale, FAD = McMaster Family Assessment Device, BDI = Beck Depression Inventory, BCFPI = Brief Child and Family Phone Interview, SSSC = The Social Support Scale for Children, CES-D = Centre for Epidemiological Studies Depression Scale, PSOC = Parental Sense of Competence Scale, MSI-R = Marital Satisfaction Inventory - Revised, CRPBI = Children's Report of Parental Behaviour Inventory, CFI = Camberwell Family Interview, GHQ = General Health Questionnaire, EPASE = Edinburgh Parental Attitude Scale to Epilepsy, SSSI = Social Stress and Supports Interview, HADS = Hospital Anxiety and Depression Scale, FES = Family Environment Scale, FRI = Family Relations Index, ICQ-P = Illness Cognition Questionnaire-Parent, PSS = Parent Stigma Scale[b], DFSS = Dunst Family Support Scale, GAD-7 = Generalised Anxiety Disorder, QIDS-SR16 = Quick Inventory of Depressive Symptomatology, PRPC = Parent Report of Psychosocial Care, PSI = Parenting Stress Index, PACIQ-R = Parent-Child Interaction Questionnaire Revised, SDS = Self-Rating Depression Scale, FACES = Family Adaptability and Cohesion Evaluation Scales, IPOV = Interactional Problem Solving Questionnaire, FAM III = Family Assessment Measure III, FFS-Seizure = Family Stress Scale-Seizure, PRPC, CBCL = Child Behaviour Checklist (YSR = Youth Self-Report), SBS (teacher rated) = Student Behaviour Survey, Vineland-II = Vineland Adaptive Behaviour Scales – Interview Edition, QOLCE = Quality of Life in Childhood Epilepsy, BASC-2 = Behavioural Assessment Symptom for Children – 2nd Edition, RSS = Rosenberg Self-Esteem Scale, PSS = Perceived Stress Scale[a], CSCS = Piers-Harris Children's Self-Concept Scale, CHEQOL = Childhood Epilepsy Quality of Life scale, PAAS = Pre-Adolescent Adjustment Scale (PAAS), CPMS = Childhood Psychopathology Measurement Schedule, PTRBS = Parental and Teacher Behavioural Scales, HSPQ = Harter Self-Perception Questionnaire, DCGM = Disabkids Chronic Generic Measure, RCADS = Revised Childhood Anxiety and Depression Scale, SDQ = Strengths and Difficulties Questionnaire, CPRS = Conners Parental Rating Scale, SSRS = Social Skills Rating System, ICIS = Impact of Childhood Illness Scale, PEDS-QL = Pediatric Quality of Life Inventory,

3.3 Outcome of studies: Associations between family factors and psychosocial outcomes in childhood epilepsy

The main aim of this review was to synthesise the relationships between various psychological family factors and psychosocial outcomes in childhood epilepsy, the results of which are visually presented in supplementary Table 1, Appendix 4. For ease of interpretation, these results will be presented by 1) family environment factors: functioning; stress/demands; resources; resilience, and 2) parent/caregiver factors: parental mental health; parental worry about epilepsy; parent-child relationship; parenting style; perceived stigma; and quality of life. A full list of psychological family factors and psychosocial outcome measures and their descriptions can be found in Appendix 5. A further aim of this review was to assess the relative importance of family factors in comparison to epilepsy factors. Overall in 9/30 studies this differential relationship could not be assessed, in 1 of the 30 studies neither family nor epilepsy factors reached statistical significance, 5/30 studies found stronger associations for epilepsy factors over family factors, and 15/30 studies documented stronger associations for family factors over epilepsy factors. These are presented in Table 2 (the factor with most strongly associated is indicated per study in the table) and are integrated throughout sections 3.3.1 and 3.3.2.

Table 2: Factor most strongly associated with child outcome

Study (main author)	Epilepsy factor	Family Factor	Relationship could not be assessed
Austin [38]			✓
Austin [39]			✓
Austin [40]	✓		
Baum [41]			✓
Carson [42]		✓	
Chapieski [43]			✓
Chew [44]			✓
Conway [45]		✓	
Ekinci [46]		✓	
Ferro [4]		✓	
HERQULES [32–36]		✓	
Han [47]		✓	
Hoare [48]			✓
Hodes [49]			✓
Kerne [50]		✓	
Li [51]			✓
Malhi [52]		✓	
McCusker [25]	✓		
McLaughlin [53]	✓		
Mendes [20]		✓	
Moreira[a] [54]	-	-	-
Pal [55]		✓	
Puka [56]		✓	
Ramsey [8]	✓		
Rodenburg [9]		✓	
Thornton [57]			✓
Tse [58]		✓	
Williams [59]	✓		
Wu [60]		✓	
Xu [37]		✓	

[a]Neither epilepsy nor family factors were significantly associated with outcomes

3.3.1 Family environment factors

3.3.1.1 Family functioning

Findings from 8/9 studies investigating the relationship between family functioning and emotional/behavioural adjustment suggested that lower levels of family functioning were related to increased difficulties with emotional/behavioural adjustment, with small – medium effect sizes across studies at univariate level [8,9,25,37,39,47,52,56]. However, results from two of these studies [39,47] highlighted that family functioning did not contribute any unique variance to emotional and behavioural adjustment factors at multivariate

level. One study [8] did not find any significant association between family functioning and emotional/behavioural adjustment over a 25-month period. Furthermore, another study [56] broke up the construct of emotional and behavioural adjustment by reporting on both depression and anxiety symptoms in children and adolescents individually. They found that at multivariate level, family functioning *was not associated with child or adolescent anxiety symptoms*, nor was it associated with *adolescent depressive symptoms*, however, was significantly related to *child depressive symptoms*. Two studies [25,52] added to overall findings by highlighting that higher levels of cohesion were also related to lower levels of emotional and behavioural difficulties.

Three out of four studies [20,33–36,45] investigating family functioning and child quality of life found that better family functioning predicted better quality of life, with medium effect. However, one study [8] found that family functioning only predicted the social interaction subscale of the QOLCE, and did not significantly contribute to any other subscale, as measured over a 25-month period. Rather, they referenced epilepsy factors as primary predictors of quality of life.

Two studies measured family functioning and social competence [57,58]. Both studies concluded the extent of family dysfunction was related to lower levels of social competence with medium effect. The latter study [58] found that family functioning was more highly correlated with social competence than neurological variables. Two studies [46,52] evaluated family functioning and self-concept and highlighted that lower family functioning was associated with

lower self-concept, and both reported medium effect sizes for same and greater significance than epilepsy variables.

3.3.1.2 Family stress and demands

Six studies investigated the relationship between family stress and psychosocial outcomes (emotional and behavioural adjustment, quality of life, and adaptive functioning reported inconsistent findings with regard to its level of impact on outcomes. Three studies [33,34,36,45,60] concluded that increased family stress and demands were related to poorer quality of life in children and young people with epilepsy, with one study [60] realising this impact consistently over a two-year period. Another study [43] found that higher levels of family stress negatively impacted the communication domain of adaptive functioning but was not significantly associated with daily living or social skills domains. Furthermore, no significant associations between family stress and child and adolescent anxiety symptoms and child depressive symptoms were found in another study [56]. They did find a significant relationship between adolescent depressive symptoms and family stress. In contrast [40] found that higher family stress was associated with emotional/behavioural at univariate and multivariate levels.

3.3.1.3 Family resources

Seven studies looked at the association between family resources (operationalised as resources available to aid families in times of distress, e.g. level of mastery, esteem, social support, etc.) and child psychosocial outcomes, with one study investigating the impact as a moderator of outcome. Greater

family resources were associated with better quality of life [32,34,36,45], with one study illustrating that family resources also moderated the impact of parental depression on child quality of life [32]. Lower family resources were associated with poorer emotional and behavioural adjustment [38,39] and adaptive skills (daily living skills) [43] with one study [39] reporting the relationship to be consistent at both baseline and two years later. However, one study [56] found that while family resources significantly predicted child and adolescent depressive symptoms and adolescent anxiety symptoms at multivariate level, and over and above that of epilepsy variables, it was not associated with child anxiety at any level. An investigation into the moderating effect of family resources on infant temperament and child behaviour problems found that children who had been identified with a 'difficult' temperament as infants, and who lived in homes that had fewer resources available to them, tended to have more emotional/behavioural difficulties than those children who were associated with 'easy' temperaments [41]. Moreover, they also found CWE with 'difficult' temperaments that lived in families with higher levels of resources were not significantly at risk for emotional and behavioural difficulties.

Parental social support was specifically measured in three studies. Outcomes measured included child emotional/behavioural adjustment [40,55] and quality of life [4]. Studies evaluating social support and emotional/behavioural adjustment found contrasting results. While one study [40] found that lower levels of family social support were the strongest family predictors of behavioural difficulties (second to female gender and seizure frequency), higher satisfaction with social support was also associated with more behavioural

difficulties in another study [56]. Findings with regard to quality of life [4] were more in line with Austin and colleagues study [40] in that higher parental social support was found to protect against low quality of life.

3.3.1.4 Family resilience

One study [44] looked at the relationship between family resilience and self-esteem in young people with epilepsy and found that higher levels of family resilience were related to higher self-esteem, explaining 67% of the variance.

3.3.2 Parent/caregiver factors

3.3.2.1 Parental mental health

The relationship between parental mental health and child outcomes was measured in 11 out of 30 studies. Overall parental mental health status was moderately positively correlated with child emotional/behavioural difficulties [48,49]. Parental depression was related to higher levels of difficulty in emotional and behavioural adjustment [9,47,56] with medium effect, and was the strongest predictor of such, when controlling for epilepsy variables[9,47]. Similarly, higher levels of parental depression were associated with, and predictive of, lower self-concept scores [46], over and above epilepsy variables. One study [56] did not find a significant association between parental depression and child anxiety symptoms but did for child depression and adolescent depression and anxiety. These associations held true for multivariate level also and highlighted the parental anxiety levels were the strongest predictor of adolescent mental health, and parental depression was the strongest predictor of child depression, independent of epilepsy variables.

Increased levels of parental depression and anxiety were found to consistently negatively impact on child quality of life in 5/5 studies [4,32–36,45,51,59] with one study [4] identifying parental depression as the most robust predictor of child health related quality of life in a 28-month period. This study did find that clinical factors (number of anti-epileptic medications, seizure severity) also predicted quality of life over time but had a lesser impact than parental depression. Similarly, high levels of parental anxiety were negatively correlated with child quality of life [51,59] portraying a medium effect size and explaining 50% of the variance at multivariable level [59]. One study noted that over a two-year period, the impact of parental depression was partially mediated through family functioning and family stress, and moderated by family resources [32].

3.3.2.2 Parental worry about epilepsy

Eight studies [8,39,42,43,48,50,53,60] measured parental anxiety/worry about epilepsy, with 7/8 studies reporting a significant association with child outcomes.

Specifically, one study [53] found that more parental thoughts of helplessness were significantly negatively associated with decreased child quality of life (relationship was significant also at multivariable level) and another study [60] found parental fears and concerns about epilepsy impacted overall quality of life over a two-year period. Such associations were not reported for seizure variables. Furthermore, parental worry impacted emotional/ behavioural adjustment over a two-year period in one study [39], and parental worry and

attitudes about epilepsy were also associated with child behavioural difficulties in a cross-sectional study [48]. Furthermore, a study measuring social competence found that higher parental anxiety about epilepsy was moderately related to poorer child social competence rated by parents and teachers, specifically poorer social skills and greater social problems, and was the strongest predictor of social competence over epilepsy variables [42]. Both studies [43,50] considering adaptive functioning found small, significant relationship with daily living skills, socialisation and communication at both univariable and multivariable level. The former of these studies [43] further found the negative relationship between parental anxiety about epilepsy and the communication and socialisation domains to be significant one year later at multivariable level. However, one study [8] found that parental worry made no significant contribution to child overall quality of life, nor was it significantly predictive of any of the individual subscales of quality of life.

3.3.2.3 Parenting (including parent-child relationship and marital satisfaction)

Parent-child interaction/relationship were measured in four studies. One study [49] investigated maternal expressed emotion (critical comments, emotional over-involvement, warmth, and positive remarks) as a measure of the parent-child relationship and found that critical comments were significantly associated with child total behavioural difficulties. These authors also assessed expressed emotion with child self-concept but did not find any significant associations with overall scores. Another study [47] assessed parent-child relationships and found

that high levels of perceived parental rejection and an over-controlling parenting style, positively correlated with child emotional and behavioural difficulties.

Stressful parent-child relationships were found to negatively impact on child quality of life over a two-year period, although decreased in impact over time, in one study [60]. Further, negative parent-child relationships correlated with increased child emotional and behavioural difficulties, while positive parent-child relationships served to protect against them, in another study [9].

Two studies [9,47] measured the relationship between parenting sense of competence and child emotional and behavioural adjustment. Both found a small association between the two variables, however the former [9] found significance for internalising symptoms only.

Finally, higher levels of marital distress and more conflict over childrearing were linked to child emotional and behavioural difficulties, although again the association was small in one study [47], and marital dissatisfaction was linked to behavioural difficulties in another [9].

3.3.2.4 Perceived stigma

Perceived stigma and its effect on child quality of life was measured in two studies. Both studies [20,60] found that when levels of perceived stigma were higher, quality of life was lower for CWE with the latter study illustrating that parental perceived stigma consistently impacted on child quality of life over a two-year period.

3.3.2.5 Parental quality of life

One study [20] showed that better parental quality of life was associated with better child quality of life, with moderate effect, however this was contrasted by another study [54], which did not find any significant association.

4. Discussion

The primary aim of this review was to synthesise the findings from studies evaluating the relationship between psychological family factors and psychosocial outcomes in childhood epilepsy. Looking at the evidence reviewed from the studies as a whole, it would certainly appear that psychological family factors – both environmental and parent/caregiver - are undeniably important in considering outcomes for CWE, with 29/30 studies (one contrasting study – [54]) reporting at least a small association between at least one family factor measured and child outcomes. The most consistent findings were in relation to parent/caregiver factors and child/adolescent outcomes, whereby all studies evaluated (excluding one – [8]) found significant moderate associations at both univariate and multivariate level (where analysed).

Notwithstanding the impact of epilepsy on both the child and family themselves, it appears that how families function – how they relate to each other, cope with stress, use available resources, and experience their own mental health, among other factors – is actually as, if not more, important for child outcomes than living with epilepsy. Plainly, this suggests that how a child with epilepsy functions, behaves and experiences themselves, and their world, is in some cases

influenced more by the families they reside in, than it is living with a chronic illness. At a broader level, this also means that early identification of the family related factors that are amenable to change and the input of appropriate supports for families who are at risk of dysfunction, could act as a protective buffer for the children and adolescents themselves, and possibly prevent them from experiencing psychosocial maladjustment that is often comorbid with epilepsy.

Variability in some findings may be the result of the range of outcome measures utilised, or a feature of the specific samples. For example, one study [56] did not find significant relationships between family factors and child anxiety. In this case, it can be noted that the presenting levels of anxiety of children in their study were not excessively high and so could account for this finding. It should also be noted here that epilepsy factors were not associated with child anxiety either, which further supports this hypothesis. Another study [55] found that increased parent social support actually contributed to higher emotional behavioural difficulties. This result appears counter-intuitive at face value and may be due to the construct of social support in Eastern versus Western countries, whereby many wider family members are involved in the care of the child and perhaps may not always be viewed as positively due to differing points of view. It must also be noted here that this study obtained the lowest quality index rating in the paper and so may account for this result. Furthermore, one study [8] did not find any significant relationship between family functioning and overall quality of life for CWE, however, they did find a predictive relationship on the social subscales of quality of life. This may perhaps be reflective of the sense of stigma of epilepsy that some families experience and

so not wanting their child to be seen as different socially. Alternatively, it could also be the result of parental fear as to what will happen to their child if they have a seizure in their absence and so their opportunities for social engagement are restricted [61]. Lastly, two studies measuring relationship between parental quality of life and child quality of life found contrasting results [20,54]. In both studies quality of life was measured by child responses, however the latter study used a more general measure of subjective health and well-being and the former measured the impact of having a chronic illness. Both studies were carried out on a Portuguese sample of CWE with similar levels of epilepsy severity.

Looking more at the specific findings, studies investigating family factors and child emotional and behavioural adjustment, quality of life, social competence, self-concept/esteem, and adaptive functioning found that poorer outcomes were associated with: higher levels of family dysfunction; higher levels of family stress (child depressive symptoms only); lower family resources; decreased parental mental health; increased parental worry; poorer parent-child relationships; increased parenting difficulties; higher perceived stigma; and lower parent perceived competence. Better child and adolescent outcomes were associated with higher parental quality of life, higher levels of family resilience, better family functioning, and more positive parent child relationships. Certainly, these findings are imperative to the scaffolding of families in promoting more positive outcomes for the future.

It must be acknowledged that although this review did not investigate the impact of structural family factors (e.g. socioeconomic status, unemployment, etc.) on

outcomes for CWE, it is likely that these factors are also influential. It may indeed be that such structural difficulties also influence outcomes for CWE directly, or by impacting on psychological family factors which then influence child outcomes. In considering this, such families that indicate more risk factors through both structural and psychological family vulnerabilities require a more multi-system intervention.

4.1 Limitations

Firstly, findings should be interpreted with caution. This review focused on literature that was for the most part cross-sectional in nature. Thus, the direction of causality between factors is difficult to evaluate, and as such, cannot be determined conclusively in these studies. It is possible that child outcomes are having a similar effect on family factors. Certainly, this gives us a viewpoint of the associations between these variables, however it does so only at a given time point. That said, seven studies reported prospective data, which continued to support the relationships found at baseline, suggesting consistency in these findings. Still, more longitudinal research is needed to investigate the transactional nature of these factors and infer more conclusive results over time.

Furthermore, the majority of studies (n = 17) included only parent—proxy reports of child outcomes, with a further nine including child self-reports – the 'gold-standard' for reporting. However, research has highlighted that studies which reported outcomes rated by both children and parents had similar scores [4]. This suggests that the use of parent proxy reports are valid representations of child experiences, which bodes well for the current review. Six out of 27

studies also included reports from teachers, thereby adding a non-biased measure of functioning to the parent/caregiver. Results of both parent and teacher reports were generally consistent, with teachers reported similar patterns of difficulties to parents, however to a lesser degree. Studies should aim to incorporate viewpoints from parents, children and teachers where possible in order to produce the most accurate picture of life for the young person.

Measures of outcome used were based on population norms in most studies, particularly when referencing emotional and behavioural adjustment. This may mean that some aspects of these scales may be unduly represented, for example somatic symptoms, in this population group. Positively however, measures of quality of life appeared to be more directed specifically for childhood epilepsy. This potentially presents an area of future research – the development of reliable and valid measures of psychological and behavioural outcome based on the presentation and specific needs of CWE.

Lastly, although the majority of studies excluded children with developmental/neurological disorders, such as Intellectual Disability (ID), Attention Deficit Hyperactivity Disorder (ADHD) and Autism Spectrum Disorder (ASD), in their samples, there were a number of studies which included/did not account for such difficulties. Specifically, 3/30 studies included children with developmental/neurological disorders [45,53,59], and 2/30 studies did not clarify such inclusion/exclusion criteria [37,51] in their studies regarding family factors and outcomes for children with epilepsy. This is important to consider as these conditions are commonly co-morbid with epilepsy and are

complex in their own right, and, as such may influence child and family outcomes independent of epilepsy needs.

4.2 Clinical Implications

Certainly, consideration of psychological family factors is necessary in understanding psychosocial development in childhood epilepsy. This is positive in thinking about preventative measures to promote healthy adjustment in children with epilepsy, as these factors are largely amenable to change through psychological intervention. The current review suggests particular consideration be given to parent/caregiver factors, which reported the most consistent associations with child outcomes. What is more is that in over half the studies reviewed, family factors were more strongly associated with child psychosocial outcomes than epilepsy factors. Certainly, there is much evidence to support that epilepsy factors, such as the amount and type of medication the child takes, the type of epilepsy, and the frequency of seizures all do understandably take their toll on children's quality of life and overall outcome. However, this research suggests that overall outcome could be within the control of the families themselves. This can give hope to families who receive new epilepsy diagnoses in that they can take proactive measures to prevent their child experiencing other additional difficulties where possible, and in doing so, take some control back from a disease that is so unpredictable by nature. It also gives hope to families whose child is experiencing severe epilepsy that they can be proactive in their child's care and endeavour to achieve positive change. This view was supported by a systematic review [16] which concluded that although clinical factors are

undoubtedly are very stressful to manage and cope with, it was the family psychosocial factors that appeared to determine parents of CWE's outcomes.

Thus, providing front-line clinicians with appropriate training and tools for assessment and brief intervention, with at-risk families, could reduce the negative psychosocial impact that some families and CWE experience. Furthermore, bolstering the resilience and resources that many families who are functioning well already hold, and equipping them with knowledge and skills about how to stay healthy themselves, communicate effectively and problem solve difficulties will have a positive impact in overall child psychosocial adjustment. This type of family-centred care has been documented in the broader literature for childhood chronic illness, and has been shown to be associated with reduced family stress and better child outcomes [21]. Moreover, a recent pilot intervention which looked at the feasibility and acceptability of a one-day parent intervention for parents of CWE has yielded positive results [62] and provides a promising outlook for future interventions to support healthy adjustment in CWE.

4.3 Conclusion

Over the past 30 years there has been increasing interest into the relationship between psychological family factors and psychosocial outcomes in paediatric epilepsy. Yet to date, no systematic review has been undertaken to formally analyse this overall relationship, despite the fact that pertinent research in the field continues to grow. From the evidence reviewed, it is clear that psychological family factors are important to consider when aiming to promote

healthy psychosocial development in childhood epilepsy. Specifically, parent/caregiver factors emerged as those most consistently associated with child outcomes, seconded by other family factors which have been most heavily researched (family resources, stress, functioning, etc.). The finding that family factors were more significantly associated with child psychosocial outcomes in 15/21 studies (where this relationship could be assessed) than epilepsy factors, is indeed positive as it identifies avenues for support that are amenable to positive change. Training front-line clinicians in brief interventions and enabling them to identify families who may be at risk of difficulties, will likely have a preventative effect on the inevitable stresses that life brings, both with epilepsy and without.

Disclosure of conflicts of interest

None of the authors had conflicts of interest to disclose.

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Appendix 1: Author guidelines for submission to Epilepsy and Behavior journal

(https://www.elsevier.com/wps/find/journaldescription.cws_home/622822?generatepdf=true)

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This journal operates a single blind review process. All contributions will be initially assessed by the editor for suitability for the journal. Papers deemed suitable are then typically sent to a minimum of one independent expert reviewer to assess the scientific quality of the paper. The Editor is responsible for the final decision regarding acceptance or rejection of articles. The Editor's decision is final. More information on types of peer review.

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To avoid unnecessary errors you are strongly advised to use the 'spell-check' and 'grammar-check' functions of your word processor.

Article structure

Subdivision - numbered sections

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

Introduction

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

Material and methods

Provide sufficient details to allow the work to be reproduced by an independent researcher. Methods that are already published should be summarized, and indicated by a reference. If quoting directly from a previously published method, use quotation marks and also cite the source. Any modifications to existing methods should also be described.

Results

Results should be clear and concise.

Discussion

The **Discussion** section should explore the significance of the results of the work, not repeat them. **Results** and **Discussion** should be separate and may be organized into subheadings. Avoid extensive citations and discussion of published literature.

Conclusions

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

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Although a graphical abstract is optional, its use is encouraged as it draws more attention to the online article. The graphical abstract should summarize the contents of the article in a concise, pictorial form designed to capture the attention of a wide readership. Graphical abstracts should be submitted as a separate file in the online submission system. Image size: Please provide an image with a minimum of 531 × 1328 pixels (h × w) or proportionally more. The image should be readable at a size of 5 × 13 cm using a regular screen resolution of 96 dpi. Preferred file types: TIFF, EPS, PDF or MS Office files. You can view Example Graphical Abstracts on our information site.
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Immediately after the abstract, provide a maximum of 6 keywords, using American spelling and avoiding general and plural terms and multiple concepts (avoid, for example, 'and', 'of'). Be sparing with abbreviations: only abbreviations firmly established in the field may be eligible. These keywords will be used for indexing purposes.

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

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List funding sources in this standard way to facilitate compliance to funder's requirements:

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Ensure that each illustration has a caption. Supply captions separately, not attached to the figure. A caption should comprise a brief title (**not** on the figure itself) and a description of the illustration. Keep text in the illustrations themselves to a minimum but explain all symbols and abbreviations used.

Tables

Please submit tables as editable text and not as images. Tables can be placed either next to the relevant text in the article, or on separate page(s) at the end. Number tables consecutively in accordance with their appearance in the text and place any table notes below the table body. Be sparing in the use of tables and ensure that the data presented in them do not duplicate results described elsewhere in the article. Please avoid using vertical rules and shading in table cells.

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Please ensure that every reference cited in the text is also present in the reference list (and vice versa). Any references cited in the abstract must be given in full. Unpublished results and personal communications are not recommended in the reference list, but may be mentioned in the text. If these references are included in the reference list they should follow the standard reference style of the journal and should include a substitution of the publication date with either 'Unpublished results' or 'Personal communication'. Citation of a reference as 'in press' implies that the item has been accepted for publication.

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As a minimum, the full URL should be given and the date when the reference was last accessed. Any further information, if known (DOI, author names, dates, reference to a source publication, etc.), should also be given. Web references can be listed separately (e.g., after the reference list) under a different heading if desired, or can be included in the reference list.

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[4] Mettam GR, Adams LB. How to prepare an electronic version of your article. In: Jones BS, Smith RZ, editors. Introduction to the electronic age, New York: E-Publishing Inc; 2009, p. 281–304.
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[5] Cancer Research UK. Cancer statistics reports for the UK, http://www.cancerresearchuk.org/aboutcancer/statistics/cancerstatsreport/; 2003 [accessed 13 March 2003].
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Appendix 2: Modified Downs & Black Quality Index

		Yes	No	Unclear
Reporting				
1.	Is the hypothesis/objective of the study clearly described?	1	0	0
2.	Are the main outcomes to be measured clearly described in the Introduction or Methods section?	1	0	0
3.	Are the characteristics of the patients included in the study clearly described?	1	0	0
4.	Are the main findings of the study clearly described?	1	0	0
5.	Does the study provide estimates of the random variability in the data for the main outcome?	1	0	0
6.	Have actual probability values [or confidence intervals] been reported for the main outcomes except where the probability value is less than 0.001?	1	0	0
7.	Is the response rate clearly described?	1	0	0
External Validity				
8.	Were the patients asked to participate in the study representative of the entire population from which they were recruited?	1	0	0
9.	Were patients who were prepared to participate representative of the entire population from which they were recruited?	1	0	0
10.	Were the staff, places, and facilities where the patients were studied, representative of the treatment the majority of patients receive?	1	0	0
Interval Validity				
11.	If any of the results of the study were based on "data dredging," was this made clear?	1	0	0
12.	Were the statistical tests used to assess the main outcomes appropriate?	1	0	0
13.	Were the main outcome measures used valid and reliable?	1	0	0
14.	Was there adequate adjustment in the analyses from which the main results were drawn	1	0	0
Power				
15.	Did the study provide a sample size or power calculation to detect important effects where the probability value for a difference being due to chance is less than 0.05?	1	0	0

Note: The original 27-item Quality Index was modified to exclude assessment of items related specifically to intervention studies, including randomization, blinding, withdrawals and drop-outs, and intervention integrity, reducing it to 15 items

Appendix 3: Summary of the modified Downs & Black quality assessment

Study (main author)	Total score (max= 15)	Reporting Quality (max = 7)	External Validity (max=3)	Internal Validity (max = 4)	Power Calculation (max = 1)
Austin [38]	11	6	2	3	0
Austin [39]	12	6	2	4	0
Austin[40]	11	5	3	3	0
Baum [41]	13	6	3	4	0
Carson [42]	10	5	1	4	0
Chapieski [43]	13	7	2	4	0
Chew [44]	14	7	3	4	0
Conway [45]	13	7	2	4	0
Ekinici [46]	15	7	3	4	1
Ferro [32]	14	7	3	4	0
Ferro [33]	13	7	3	3	0
Ferro [4]	14	7	3	4	0
Goodwin [34]	13	7	3	3	0
Han [47]	12	6	2	4	0
Hoare [48]	7	3	2	2	0
Hodes [49]	14	7	3	4	0
Kerne [50]	9	4	1	4	0

Li [51]	7	3	1	3	0
Malhi [52]	11	5	2	4	0
McCusker [25]	12	5	3	4	0
McLaughlin [53]	11	6	1	4	0
Mendes [20]	14	7	3	4	0
Moreira [54]	12	6	2	4	0
Pal [55]	6	4	0	2	0
Puka [56]	14	7	2	4	0
Ramsey [8]	14	7	3	4	0
Rodenburg [9]	14	7	3	4	0
Sajobi [35]	14	7	3	4	0
Speechley [36]	14	7	3	4	0
Thornton [57]	13	7	2	4	0
Tse [58]	14	7	3	4	0
Williams [59]	10	4	2	4	0
Wu [60]	14	7	3	4	0
Xu [37]	13	7	2	4	0
Mean (SD)	12 (2.3)	6 (1.3)	2.3 (.8)	3.7 (.6)	.03 (.2)

Appendix 4: Supplementary Table 1

Supplementary Table 1: Summary of the relationships between family factors and psychosocial outcomes in children with epilepsy

Note: All relationships are in the expected direction and at the univariable level unless otherwise stated

		Child Psychosocial Outcome									
		Emotional/ Behavioural Adjustment	Quality of Life	Social Competence			Self-Concept	Self-esteem	Adaptive Functioning		
				Social skills	Social problems	School			Communication	Daily Living Skills	Social
Austin [38]	Family Functioning	*M									
	Family Resources	*M									
Austin [39]	Family Functioning										
	Parental Worry										
	Family Resources										
Austin [40]	Family Stress	*M									
	Family Resources (social support)	*M									
Baum [41]	Low Family Resources[A]	***									
	High Family Resources[A]	-									
Carson [42]	Parental Worry about Epilepsy			**	*						
Chapieski [43]	Parental Worry about Epilepsy								***M	*	*M
	Stressful Life Events								***	-	-
	Family Resources								-	*** M	-
	Parenting Style								-	***	***
Chew [44]	Family Resilience						**				
Conway [45]	Family Functioning		**								
	Family Resources		**M								
	Stressful Life Events		**								
	Parental Mental Health		**								
Ekinici [46]	Family Functioning						***M				
	Parental Mental Health						***M				
Ferro [4]	Parental Mental Health		*M								
	Social Support		*M								
HERQULES [32–36]	Family Functioning		*								
	Family Resources		*								
	Stressful Life Events		*								

Supplementary Table 1: Summary of the relationships between family factors and psychosocial outcomes in children with epilepsy

Note: All relationships are in the expected direction and at the univariable level unless otherwise stated

			Child Psychosocial Outcome										
			Emotional/ Behavioural Adjustment	Quality of Life	Social Competence			Self-Concept	Self-esteem	Adaptive Functioning		
					Social skills	Social problems	School			Communication	Daily Living Skills	Social
	Parental Mental Health			*								
Han [47]	Family Functioning		***									
	Parenting Competence		***									
	Parental Mental Health		*									
	Parenting Conflict		**									
	Marital Satisfaction		***									
	Parent child relationship	Rejection	***									
		Overcontrol	*									
Hoare [48]	Parental Mental Health		**									
	Parental Worry about Epilepsy		**									
	Parental Mental Health		***									
Hodes [49]	Parent-Child Relationship	Critical Comment	***									
		Emotional over - involvement	-									
		Warmth	-									
		Positive remarks	-									
Kerne [50]	Parental Worry about Epilepsy									*M	*M	**M
Li [51]	Parental Mental Health			*								
Malhi [52]	Family Functioning		**					***				
McCusker [25]	Family Functioning	Cohesion	**									
		Conflict	**									
McLaughlin [53]	Parental Coping (helplessness)			**M								

Supplementary Table 1: Summary of the relationships between family factors and psychosocial outcomes in children with epilepsy

Note: All relationships are in the expected direction and at the univariable level unless otherwise stated

			Child Psychosocial Outcome													
			Emotional/ Behavioural Adjustment				Quality of Life	Social Competence			Self-Concept	Self-esteem	Adaptive Functioning		
								Social skills	Social problems	School			Communication	Daily Living Skills	Social
Mendes [20]	Family Functioning						**								
	Parental QoL						**								
	Stigma						**								
Moreira [54]	Parental QoL						-								
Pal [55]	Social Support[B]				***										
Puka [56]			Child Anxiety	Child Depression	Adolescent Anxiety	Adolescent Depression									
	Family Functioning		-	***	-	-									
	Family Resources		-	***	***	***									
	Stressful Life Events		-	-	-	***									
	Parental Mental Health	Anxiety	-	-	**[M]	***[M]									
		Depression	-	***[M]	***	***[M]									
Ramsey [8]	Parental Worry about Epilepsy				-		-								
	Family Functioning				-		-								
Rodenburg [9]	Family Functioning				***(internalising) **(externalising)										
	Parental Mental Health				**										
	Parent-Child Relationship				*										
	Marital Relationship				**(externalising)										
	Parenting Competence				***										
Thornton [57]	Family Functioning				*					**					
Tse [58]	Family Functioning									*					
Williams [59]	Parental Mental Health						**[M]								
Wu [60]	Family Stress						*[M]								
	Fears and Concerns						**[M]								
	Stigma						-								

Supplementary Table 1: Summary of the relationships between family factors and psychosocial outcomes in children with epilepsy

Note: All relationships are in the expected direction and at the univariable level unless otherwise stated

		Child Psychosocial Outcome									
		Emotional/ Behavioural Adjustment	Quality of Life	Social Competence			Self-Concept	Self-esteem	Adaptive Functioning		
				Social skills	Social problems	School			Communication	Daily Living Skills	Social
Xu 2017	Parent-Child Relationship Stress		-								
	Family Functioning	***									

- = no significant relationship, [M] Results significant at multivariate level, [A]Moderator of outcome, [B]direction of significance was not expected
* $p < 0.001$
** $p < 0.01$
*** $p < 0.05$

Appendix 5: Family factor and psychosocial outcome measures and descriptions

Psychosocial measures used

Construct	Scale	Description	Study
Emotional and Behavioural Adjustment	RCADS: Revised Childhood Depression and Anxiety Scale	47-item scale Assesses symptoms of depression and anxiety in children and adolescents aged 6-years Items are scored on a 4-point scale and interpreted via T-score (M = 50, SD =10) based on the child's grade and sex to identify children in the average, borderline and clinical ranges.	Puka [56]
	Generalised Anxiety Disorder (GAD-7)	7-item scale Designed to screen for anxiety disorder in the general population Items are scored on a 4-point scale (0 = not at all sure – 4 = nearly every day), with total scores ranging from 0-21. Scores noted in the moderate to severe ranges (above 9) as indicative of risk of anxiety.	Puka [56]
	Quick Inventory of Depressive Symptomatology (QIDS-SR16)	16-item scale Designed to screen for depression in the general population. Items are rated on a 4-point scale to measure nine symptom domains (Sleep disturbance, Sad mood, Decrease/increase in appetite/weight, Concentration, Self-criticism, Suicidal ideation, Interest, Energy/fatigue, Psychomotor agitation/retardation). Scores range from 0-27, higher scores are indicative of greater risk of depression.	Puka [56]

	The Behavioural Assessment System for Children 2nd Ed. (BASC-2)	Designed to measure behavioural and emotional difficulties	Ramsey [8]
	Child Behaviour Checklist (CBCL)	118-item scale Designed to measure children aged 6-18 years emotional and behavioural adjustment over the previous 6 months. Items are rated on a 3-point scale (0 = not true – 3 =very/often true). Scores are computed for Total Problems, Internalising Problems (withdrawals, somatic complaints and anxiety/depression), Externalising Problems (aggression, delinquent behaviour).	Austin [38] Austin [39] Austin [40] Baum [41] Carson [42] Han [47] McCusker [25] Rodenburg [9] Tse [58]
	Social Problems and Social Skills subscales of the Student Behaviour Survey (SBS)	102-item scale Standardised measure of teacher report of behavioural functioning.	Carson [42]
	Kovac Children's Depression Inventory (CDI)	10-item scale Designed to measure depressed mood in children across five domains: negative mood, ineffectiveness, negative self-esteem, interpersonal problems, and anhedonia. Items are scored on a 3-point scale from 0 (not at all true) to 2 (very true), with higher scores reflecting more depressive symptoms.	Ferro [4]
	Conners Rating Scale Revised (CRS-R)	28-item scale Teacher rating scale used to screen for behavioural difficulties in children. The scale provides a total score for three subscales: attention deficit; hyperactivity; conduct problems.	Ekinci [46] Pal [55]
	Turgay DSM-IV Disruptive Behavior Disorders rating Scale (T-DSM-	Based on DSM-IV diagnostic criteria and assessed hyperactivity, impulsivity, inattention, opposition-defiance, and conduct disorder.	Ekinci [46]

	IV-S; parent and teacher forms)	Items are scored on a 4-point scale from 0 (not at all) to 3 (very much).	
	Strengths and Difficulties Questionnaire (SDQ)	25-item measure Designed to measure psychological adjustment in children and adolescents. Items are answered on a 3-point scale (0 = not true – 2 = certainly true), with higher scores reflecting greater difficulties.	Moreira [54] Xu [37]
	Disabkids Chronic Generic Measure (DCGM)	12-item scale Measures the perceived impact of the chronic illness and treatments on the individual's life. Items are rated on a 5-point scale ranging from 1 (never/not at all) to 5 (always/extremely). Higher scores are reflective of greater health related quality of life.	Mendes [20]
	Childhood Psychopathology Measurement Schedule (CPMS)	75-item scale Measure of emotional and behavioural adjustment based on a Hindi adaptation of the CBCL.	Mahli [52]
	Parental and Teacher Rutter Behavioural Scales (PTRBS)	Used to assess 'psychological deviance' in children. Items are scored on a 3-point scale with children scoring 13 and over on the parent scale and 9 or over on the teacher scale, at risk.	Hoare [48] Hodes [49]
	Perceived Stress Scale (PSS)	14-item scale Designed to assess young people's perceptions of stress. Items are rated on a 5-point scale (1 = never – 5 = very often), with higher scores indicating higher levels of stress.	Chew [44]
Quality of Life	Pediatric Quality of Life Inventory – Epilepsy Version (PedsQL)	23-item scale Designed to measure parental perceptions of HRQOL in children aged 2-18 years. Higher	Wu [60]

		scores indicate greater HRQOL	
	Impact of Child Neurological Disorder (ICND)	44-item scale Measures impact on functioning across four domains: behaviour, cognition, physical/neurological disability, and epilepsy). Quality of life is rated from 1-6 (1 = poor to 6 = excellent). Higher scores indicate lower quality of life	Tse [58]
	Quality of Life in Childhood Epilepsy (QOLCE) questionnaire	91-item scale Designed to measure health related quality of life in children aged 6-18 years with epilepsy. Items are rated on a 5-point scale ranging from (very often or all of the time) to (never or none of the time). Scores range from 0-100, with higher scores reflecting greater quality of life	Conway [45] HERQULES [32–36] Li [51] McLoughlin [53] Ramsey [8] Wu [60]
	Childhood Epilepsy Quality of Life scale (CHEQOL)	25-item scale Designed to assess epilepsy specific quality of life across five domains: personal/social consequences, worries and concerns, intrapersonal/emotional issues, secrecy, and quest for normality. Responses are scored from 0-100 with higher scores indicating better QoL.	Ferro [4]
	The Impact of Childhood Illness Scale (ICIS)	30-item scale Assesses quality of life in children with a chronic illness. Each item is rated as ‘never or rarely true’, ‘sometimes true’, or	Williams [59]
	KIDSCREEN-10 index	10-item index Designed to measure the general subjective health and well-being of children and adolescents aged 8-18 years. Items are scored on a 5-point scale (1 = never;	Moreira [54]

		not at all – 5 = always; extremely), with higher scores reflecting better QoL.	
Adaptive Functioning	Vineland Adaptive Behaviour Scales – Interview Edition (Vineland-II) survey form	Semi-structured interview completed with a primary caregiver to assess social and personal independence in day-to-day functioning. Items scored from 0-3 (never performs the task – usually performs the task). Provides total standard scores for functioning categorised into Communication, Socialisation and Daily Living Skills domains.	Chapieski [43] Kerne [50]
	Pre-Adolescent Adjustment Scale (PAAS)	40-item scale Assesses adjustment towards school, home, teachers, peers, and general issues. High positive scores indicate good adjustment.	Mahli [52]
	Rosenberg Self-Esteem Scale (RSS)	10-item scale Evaluates self-esteem through positive and negative perceptions of the self. Items are rated on a 4-point scale (1 = strongly disagree – 4 = strongly agree), with higher scores reflecting higher levels of self-esteem.	Chew [44]
Self-Esteem	Harter Self-Perception Questionnaire (HSPQ)	Consists of a series of statements which measures a child's view of themselves across a number of domains including scholastic competence, social acceptance, athletic competence, physical appearance, and behavioural conduct.	Hodes [49]
	Piers-Harris Children's Self-Concept Scale (CSCS)	80-item scale Designed to measure self-concept in children aged 7-18 years. Responses are recorded on a Yes/No basis, with higher scores indicating greater self-concept.	Austin [38] Ekinci [46] Mahli [52]

Social Support	The Social Support Scale for Children (SSSC)	18-item scale Measures child-reported parental, classmate and close friend social support. Respondents decide whether a statement is 'really true for me' or 'sort of true for me', with higher scores indicating more social support.	Ferro [4]
	Social Skills Rating System (SSRS)	Measures behaviours that are important for the development of social competence and adaptive functioning. It comprises of both parent and student responses.	Tse [58]

Family factor measures used

Construct	Scale	Description	Study
Family Functioning (including environment and resources)	Family Adaptability, Partnership Growth, Affective and Resolve scale (APGAR)	5-item scale Designed to measure satisfaction with family relationships Items are measured on a 5-point Likert scale ranging from 0 (hardly ever) to 4 (almost always) with higher scores indicating greater satisfaction (range 0-20)	Austin [39] Auston [38] Conway [45] Han [47] HERQULES [32–36] Puka [56] Xu [37]
	McMaster Family Assessment Device (FAD)	60-item scale Designed to evaluate family functioning. The FAD is made up of seven scales which measure Problem Solving, Communication, Roles, Affective Responsiveness, Affective Involvement, Behavior Control and General Functioning. Items are rated on a 4-point scale from 1 (strongly agree) to 4 (strongly disagree). Higher scores on the general family functioning subscale suggest a lower level of family functioning.	Ekinci [46] Ramsey [8]

	Family relations Index (FRI)	27-item scale Assesses family functioning in how family members relate to each other.	McCusker [25]
	Family Environment Scale (FES)	90-item scale Designed to measure family functioning across 10 subscales, that measure the social characteristics of the family environment.	Malhi [52] Mendes [20]
	Family Assessment Measure III	50-item scale Designed to measure overall family health and family relationships	Thornton [57] Tse [58]
	Family Adaptability and Cohesion Evaluation Scales (FACES)	13-item scale Designed to measure family adaptation. Items are rated on a 4-point scale (1 = never true – 4 = always true), with higher scores reflecting more problems with family adaptation.	Rodenburg [9]
Family Resources	Family Inventory for Resource Management (FIRM)	Designed to assess resources available to families to support them with adaptation to stressful events. Items are rated on a 4-point scale (0 = not at all – 4 = very well), with higher scores reflecting a more adaptive family environment.	Austin [38] Austin [40] Austin [39] Baum [41] Conway [45] HERQULES [32–36] Puka [56]
	Coping Resources Inventory (CRI)	60-item questionnaire Designed to assess an individual's resources for coping with stress. Resource domains include Cognitive, Social, Emotional, Spiritual/Philosophical, and Physical. Items are rated on a 4-point scale, with higher scores reflecting more coping resources.	Chapieski [43]
Family Stress and Demands	Family Inventory of Life Events and Changes (FILE)	71-item scale Designed to assess family demands in the areas of intrafamily strains, marital strains, pregnancy and childbearing strains, finance and business	Austin [40] Chapieski [43] Conway[45] HERQULES [32–36] Puka [56]

		strains, work-family transition strains, illness and family care strains, losses, transitions 'in and out' of the family, and family legal violations. Uses yes/no questions with scores ranging from 0-71. Higher scores are indicative of higher demands	
	Family Stress Scale-Seizure (FSS-seizure)	14-item scale Designed to assess epilepsy specific parenting stress. Higher scores are reflective of higher levels of illness-related distress.	Wu [60]
Family Resilience	Family Resilience Assessment Scale (FRAS)	54-item scale Designed to assess family processes that support families' ability to cope successfully with adversity. Items are rated on a 4-point scale (1 = strongly disagree – 4 = strongly agree), with higher scores representing higher levels of family resilience.	Chew [44]
Parent-Child Relationship	Children's Report of Parental Behaviour Inventory (CRPBI)	26-item scale Designed as a measure of parent-child interaction. Items measured on a 5-point scale, with higher scores reflecting higher levels of feelings of rejection or control that the child feels toward the parent, and lower scores reflecting higher levels felt of love or autonomy.	Han [47]
	Camberwell Family Interview (CFI)	This is a semi-structured interview lasting approximately 1-2 hours. It focuses on areas of family life and the effects of the disorder on the relative. It is designed to measure expressed emotion between parents and children or between spouses. Ratings are based on a 6-point scale.	Hodes [49]
	Parenting Stress Index (PSI)	120-item scale	Rodenburg [9] Wu [60]

		Assesses the degree to which stress is related to parental functioning, the behavioural and temperamental qualities of the child and the parent-child relationship.	
	Parent-Child Interaction Questionnaire Revised (PACIQ-R)	21-item scale Measure of parent-child relationship. Items are scored on a 5-point scale (1 = does not apply to me at all – 5 = always), with higher scores indicating a more positive perception of parent-child relationship.	Rodenburg [9]
Parental Mental Health	General Health Questionnaire (GHQ)	Tool to screen for psychiatric disorders in the general population.	Hoare [48] Hodes [49]
	Beck Depression Inventory 2nd Edition (BDI-II)	21-item scale Designed as a screening measure of depression in adults. Items are rated on a 4-point scale, with higher scores indicating higher levels of depressive symptoms.	Ekinci [46] Han [47]
	State-Trait Anxiety Inventory for Adults (STAI)	20-item questionnaire Assesses levels of anxiety in adults. Higher scores are indicative of increased anxiety.	Ekinci [46] Williams [59]
	Quick Inventory of Depressive Symptomatology (QIDS-SR16)	16-item scale Designed to screen for depression in the general population. Items are rated on a 4-point scale to measure nine symptom domains (Sleep disturbance, Sad mood, Decrease/increase in appetite/weight, Concentration, Self-criticism, Suicidal ideation, Interest, Energy/fatigue, Psychomotor agitation/retardation). Scores range from 0-27, higher scores are indicative of greater risk of depression.	Conway [45]
	Generalised Anxiety	7-item scale	Conway [45]

	Disorder (GAD-7)	Designed to screen for anxiety disorder in the general population Items are scored on a 4-point scale (0 = not at all sure – 4 = nearly every day), with total scores ranging from 0-21. Scores noted in the moderate to severe ranges (above 9) as indicative of risk of anxiety.	
	Centre for Epidemiological Studies Depression Scale (CES-D)	20-item scale Designed to measure depressive symptoms. Items are rated on a 4-point scale, with higher scores representing more depressive symptoms.	Ferro [4] HERQULES [32–36]
	Hospital Anxiety and Depression Scale (HADS)	14-item scale Designed to measure anxiety and depression in the general population Items are scored on a 4-point scale with higher scores indicating higher symptoms of psychopathology.	Li [51]
	Illness Cognition Questionnaire Parent (ICQ-P)	18-item scale Designed to assess parent's illness cognitions about their child's illness through the categories of helplessness; acceptance; and disease benefits. Items are rated on a 4-point scale ranging from 1 (not at all) to 4 (completely).	McLaughlin [53]
	Self-Rating Depression Scale (SDS)	20-item scale Measure of feelings of depression. Items are rated on a 4-point scale (1 = seldom/never – 4 = almost always/always), with higher scores indicative of greater depressive feelings.	Rodenburg [9]
Parenting	Parental Protectiveness Scale (PPS)	12-item scale Scale produces a single-factor protectiveness score that represents parental attempts to protect their children	Chapieski [43]

		from physical harm and failure. Higher scores represent higher parental intervention	
Parental Worry	The Parental Anxiety about Epilepsy Questionnaire (PAE)	14-item questionnaire Designed to measure potential fears experienced by parents of children with epilepsy Items are rated on a Likert scale from 0-5, with higher scores indicative of greater anxiety	Carson [42] Chapieski [43] Kerne [50]
	Fears and Concerns subscale of the Parent Report of Psychosocial Care (PRPC)	5-item scale Measures the concerns, needs for care, and satisfaction with care perceived by parents of children with new-onset seizures. Higher scores indicate more fear and concern.	Austin [39] Ramsey[8] Wu [60]
	The Edinburgh Parental Attitude Scale to Epilepsy (EPASE)	45-item questionnaire Divided into 7 subsections (problems with seizures, aetiology, problems for the child now and in the future, adverse effects of drugs, parental concerns about the child, social restrictions for the child and family and adverse effects on family life), this questionnaire assesses attitudes and knowledge towards epilepsy. Each statement is rated as either ‘unlikely’, ‘possible’, or ‘probable’.	Hoare [48]
	Parental Directiveness Scale from the Parental Problem-Solving Directedness Questionnaire (PPSDQ)	6-item scale Assess parental attempts to protect their child from failure through high levels of parental involvement in school work and activities of daily living. Higher scores represent higher parental intervention	Chapieski [43]
	Parenting Sense of Competence Scale (PSOC)	16-item scale Assesses the degree to which the parent feels	Han [47]

			competent about managing the adolescent. Items are rated on a 5-point scale, higher scores reflect more confidence.	
		Conflict over childrearing and Global Distress subscales of the Marital Satisfaction Inventory-Revised (MSI-R)	10 and 22-item subscales respectively. Higher scores indicate higher levels of dissatisfaction with relationships.	Han [47]
		Satisfaction scale of the Interactional Problem-Solving Questionnaire (IPOV)	Used to measure degree of marital satisfaction. Items are rather on a 5-point scale with higher scores indicative of greater marital satisfaction.	Rodenburg [9]
	Social Support	Dunst Family Support Scale (DFSS)	Respondents rate satisfaction with 18 sources of social support on a 5-point scale.	Pal [55]
	Quality of Life	EUROHIS-QOL-8	8-item scale. Shortened version of the WHOQOL-BREF. Generates an overall score, and social, psychological, physical, and environmental subscale scores. Items are rated on a 5-point scale from 1 (not at all/very dissatisfied) to 5 (completely/very satisfied), and higher scores reflecting better quality of life.	Mendes [20] Moirera [54]
	Stigma	Parent Stigma Scale (PSS)	5-item scale Designed to measure caregivers' belief that their child is experiencing stigma or may experience stigma due to having epilepsy. Items are rated on a 5-point scale from 1 (strongly disagree) to 5 (strongly agree), with higher scores suggesting higher levels of perceived stigma.	Mendes [20] Wu [60]

Paper (2): Empirical Paper

Title: Understanding the role of family factors in predicting outcomes for childhood epilepsy

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Abbreviations: CWE, children with epilepsy

Abstract (2)

Objectives: The current study is the first to profile psychosocial outcomes for children with epilepsy attending a regional paediatric centre in Ireland. Both parent and teacher informants are utilised and disease – v- family factor associations with outcomes were examined.

Methods: Forty-eight children with epilepsy (6-16 years) and their caregivers participated in this cross-sectional survey research. Both parents and teachers reported on psychosocial adjustment, social competencies and quality of life. Exploratory bivariate correlations and then confirmatory multiple regressions were utilised to evaluate the relationship between family, child and disease related factors and psychosocial outcomes.

Results: Parental worry about their child with epilepsy, as opposed to disease severity *per se* was most strongly associated with child outcomes. Measures of disease severity (seizure frequency and number of medications) did not show any associations with child outcomes based on parent reports. However, when teacher reports were considered, the seizure frequency, as opposed to parental worry, showed more consistent associations with child psychosocial outcomes. Other family factors (resilience, cohesion and conflict) did not show any strong associations with child psychosocial outcomes.

Significance: The current study provides an insight into the profile of children and young people with epilepsy attending paediatric outpatient neurology services in Ireland. Positively, families demonstrated high levels of functioning and resilience. However, high levels of illness related parental worry were evident. Results suggest that a multi-layered approach is required for intervention, including psychoeducation

from the medical and psychological fields as well as family-based support in improving outcomes for childhood epilepsy.

Keywords: Epilepsy, Family, Caregiver, Paediatric, Psychosocial, Outcomes

1. Introduction

Although many children with epilepsy (CWE) function well day-to-day [1], a substantial percentage present with a range of psychosocial difficulties. Specifically, research has suggested an increased prevalence of adjustment difficulties with some studies suggesting that emotional difficulties (such as withdrawal, anxiety, and depression) are more preponderant than behavioural difficulties (such as acting out and conduct type difficulties) [2-5]. Likewise, CWE have been found to have decreased social skills, increased peer difficulties and more struggles with social competence [6]. Epilepsy can also impact children and young people's academic ability, and CWE have been found to have lower achievement scores than siblings, chronic illness controls and typical population children in reading, writing, maths and general information [7]. It is understandable given these noted psychosocial difficulties that CWE tend to report lower quality of life than their healthy peers [8].

Moreover, it has been posited that CWE have four times the increased risk of developing such psychosocial difficulties [9] and are reported to portray poorer outcomes than children with other chronic illnesses [10]. This evidence is supported by literature stating that children are at heightened risk for more negative outcomes when the neurological system is implicated [11-13]. Importantly, the higher prevalence of these difficulties in CWE is apparent whether or not they have achieved control over their seizures [14].

Research has thus turned to investigate what factors may act as risk or protective factors for such outcomes. The severity of epilepsy has been found to be a significant risk factor for the development of emotional and depressive difficulties, and higher

seizure frequency has been noted to have a greater impact on major aspects of child and family life (e.g. academics, parent-child relationships and family activities) than lower seizure frequency [15]. Similarly, clinical factors, such as seizure frequency, have been found to have an inverse relationship with child quality of life [16]. In contrast, other studies did not find any significant association between epilepsy variables (such as age at onset, duration of epilepsy, seizure frequency and anti-epileptic medications) and outcomes in paediatric epilepsy [17-20]. This lack of association between disease severity and psychosocial outcomes has also been found in research with other chronic illnesses [21-23]. Thus, it is clear that disease and medical factors alone cannot satisfactorily predict psychosocial outcomes for CWE.

Increasingly, emergent research evidence points towards the relatively greater importance of psychosocial factors, especially family factors, than disease related factors in understanding outcomes for CWE. For example, maternal level of education has been found to predict child quality of life [24], as well as influence child physical activity, cognition and behaviour [25]. Other studies have suggested that observation of academic achievement may be influenced by the stigma associated with epilepsy. For instance, teachers' awareness of a child's epilepsy diagnosis has been correlated with lower ratings of academic achievement, despite finding no significant relationship between the child's diagnosis and achievement on standardised tests [26].

More recently, research has begun to focus on psychological aspects of the family environment, such as family functioning, family resilience, and parental mental health, and their roles as both risk and protective factors in contributing to child outcomes in epilepsy [13,27]. Such research has also grown in the wider field of chronic illness

[28-30] though what is of particular interest and importance with epilepsy populations is that in many cases, such psychological family factors have been posited to exert more influence on child outcomes than medical variables [1,5].

However, a cautionary perspective is encouraged here, as to date such findings have been mixed and there is significant need for a systematic synthesis of such empirical literature so as to infer both causal and confirmatory relationships. For example, the association between family factors and cognitive functioning was evaluated in children with intractable epilepsy and it was concluded that epilepsy variables (e.g. age of onset, number of anti-epileptic drugs) were more important predictors of outcome than psychological family factors (family functioning, family environment) [32]. In contrast, a disorganised family environment was found to moderate the effect of neuropsychological deficits on academic achievement scores in CWE, with more disorganised homes having a more negative impact and did not find any moderating effect of seizure variables (type, duration, age of onset) [33]. More research is indicated here to examine these complex relationships.

Research has also investigated parental worry about epilepsy and outcomes for children and has noted conflicting evidence. For example while one study [34] found parental worry to predict child emotional/behavioural adjustment over a two-year period, with similar findings also noted in relation to quality of life [35], another reported no significant contribution of parental worry to child quality of life [36].

Building on this, available literature has indicated a significant negative relationship between parental depressive symptoms and child outcomes in epilepsy (see [37] for a

review). Evidence from recent prospective research concluded that not only did this relationship continue to negatively impact outcomes over a two year period [38], it was also more important in predicting outcomes than medical factors (seizure frequency and anti-epileptic medication). However, in contrast, a study assessing a similar model of factors in relation to child quality of life suggested that it was medical factors (number of anti-epileptic medications) that were more important in predicting outcomes than psychological family factors (family functioning and parental worry) [36].

Nonetheless, in thinking about families as predictors of outcome, it must be remembered that epilepsy is undoubtedly a stressful condition for the whole family [39]. As well as managing the potential presence of comorbid difficulties as outlined above, epilepsy presents as uniquely challenging due to the episodic and unpredictable complexity of seizures. Moreover, in comparison to families of healthy children, families living with childhood epilepsy are reportedly subjected to increased stress, worry and anxiety, and constraints in family life [5,40,41] as well as a number of other negative outcomes which include higher levels of marital distress [42] and decreased parental quality of life as well as increased mental health difficulties [43,44]. These are important findings in light of the above associations between family factors and child psychosocial outcomes, as effectively, as well as being negatively impacted themselves, such families may also pose as risk factors for greater difficulties in their child with epilepsy. Conversely, it would also suggest that families who display resilience when faced with such adversity could act as protective buffers for their children and promote more adaptive outcomes [45]. Thus, shifting our attention from a deficit centered approach, which focuses primarily on the absence of disease, to one

which promotes family's strengths, could help to mitigate the negative outcomes that some CWE experience.

Walsh's [46] model of family resilience provides a helpful framework for understanding why some CWE do better than others, given that such outcomes are not influenced by disease factors alone. It posits that there are key processes in how families' function that foster resilience, such as engaging in effective communication (e.g. intrafamilial and with healthcare providers) and problem-solving, ability to make meaning of their circumstances, and adopt healthy reorganizational processes so as to promote positive adjustment. Therefore, by strengthening such processes, families act as resources that can be utilised to cope and adapt successfully to adversity.

Accordingly, the current study is grounded in Walsh's principles so as to examine the relationship between a range of family processes and psychosocial outcomes for CWE. As although we may not be able to change the severity of the epilepsy, or easily effect change in structural family factors, such as socioeconomic status and family composition, we can influence the more dynamic psychological family factors through effective, strengths based psychological intervention. In line with this thinking, researchers have advocated that future research foci should deviate from comparative studies to studies that comprehensively investigate the possible family-level factors that are associated with childhood epilepsy and impairments in psychosocial outcomes more broadly [47]. Understanding these relationships, would enable the identification of a model of risk and protective factors that are amenable to change. This would thus have the effect of informing family-centred care plans and also inform the provision

of resources and the development of effective, targeted interventions in an effort to bolster the positive, healthy, psychosocial outcomes in childhood epilepsy.

The current research examines the relative associations between disease severity, family and psychosocial factors in CWE. The authors are mindful that, to date, there have been no studies profiling such outcomes in an Irish population of CWE. In addition to the generalisable significance of current study findings we wish to examine the specific outcomes evident in an Irish sample. There are two primary research questions in the current study:

1. What are the outcomes (e.g. emotional and behavioural adjustment, quality of life, social competence, family functioning, resilience and parental worry) for CWE attending paediatric services in Cork and their families?

2. Do psychological family factors (e.g. resilience, functioning, parental worry) predict psychosocial outcomes in CWE?

To date, much of the research on outcomes for CWE has (a) focused on samples with intractable epilepsy and (b) relied on parent reports. In the current study we include a wider spectrum of seizure frequency (from good to poor control) and also include independent informants of outcome – teachers.

2. Methodology

2.1. Participants and recruitment

Between February and August 2018, families of CWE (n = 159) were identified and recruited through the two of the largest city hospitals with paediatric services in the Munster region, and Epilepsy Ireland, following consultation with Clinical Nurse Specialists (CNS) in Epilepsy and Consultant Paediatric Neurologists. Families of CWE were eligible for participation subject to their children meeting the following criteria, as assessed by CNS in Epilepsy: i) a clinical diagnosis of epilepsy – the study did not differentiate children's diagnoses by seizure type ii) with active seizures (at least one seizure over the past year), iii) aged between six and 16 years of age, iv) and parents had sufficient fluency in English (so much as to give informed consent and comprehend study questionnaires). Families of CWE were excluded if their child: i) was not attending mainstream schooling – this was employed as a proxy indicator of intellectual disability ii) was diagnosed with a comorbid neurodevelopmental disorder, such as Autism Spectrum Disorder (ASD) or Attention Deficit Hyperactivity Disorder (ADHD) due to the complexity of needs often associated with such difficulties and so may have influenced child and family outcomes independent of epilepsy needs. Children with other comorbid difficulties such as asthma or physical disabilities were not excluded.

A total of 159 families were contacted and invited to participate through the Paediatric Neurology departments of the collaborating hospitals. Of these, 63 families consented to participate (40% response rate). No further information was available regarding psychological outcomes or seizure frequency for the 96 families who declined participation, however the sample were not statistically different with regard to age

and gender ($p > .05$). An additional seven families contacted the main author (EH) in response to information posted on the Epilepsy Ireland webpage regarding participation in the study.

These 70 families were subsequently sent a battery of questionnaires to be completed by parents, which included a stamped addressed envelope for ease of response. Parents were required to indicate whether the questionnaire was primarily completed by 'parent 1' or 'parent 2', although it is acknowledged that both parents may have contributed to the responses made. Therefore, as such, the data obtained may represent both parents in some households, although this was not explicitly indicated in the returned questionnaires. In total, 54 completed questionnaires were returned (77% response rate), of which six were excluded. Three were excluded due to comorbid neurodevelopmental disorder diagnoses (two ASD, one ADHD), two did not report active seizures in the previous year and one was not attending mainstream schooling. A separate questionnaire pack was also sent to the schools of the 48 families included in the study. Of these, 27 completed questionnaires were received from teachers. A summary of their demographic and clinical characteristics is presented in Table 3.

Table 3: Characteristics of families and children with epilepsy who participated in the study (n = 48), including sample with teacher-reported outcomes (n = 27)

	Overall sample		***Teacher sample***	
CWE characteristics	***N / mean ± SD***	**%**	***N / mean ± SD***	**%**
Age*	9.9 ± 3.1		8.8 ± 2.6	
Gender	48		27	
Male	19	40	12	44
Female	29	60	15	56
Age at diagnosis*	7.1 ± 5.4		5.7 ± 2.9	
Number of AED	1.5 ± .1.0		1.6 ± 1.0	
Seizure Frequency	48		27	
Previous 3 months				
No seizures	19	40	8	30
< 10	17	35	11	41
10-19	4	8	2	7
>20	8	17	6	22
Other Illness / Diagnosis in CWE[a]	47		27	
Yes	12	25	8	30
Family Characteristics				
Age				
Respondent	42.4 ± 5.5		42.1 ± 5.5	
Gender	48		27	
Male	3	6		0
Female	45	94		100
Country of birth	48		27	
Ireland	45	94	27	100
Other	3	6	0	0
Family composition	48		27	
Lone parent	14	29	8	30
Number of children in family	2.8 ± 1.5		2.9 ± 1.7	
Employment	47		27	
Respondent employed	28	59	16	59
Highest level of education	47		27	
Secondary School	13	28	9	33
Additional Training	17	36	9	33
Third level	17	36	9	33
Illness / diagnosis in either parent[b]	48		27	
Yes	17	35	11	41
Illness / diagnosis in siblings	48		27	
Yes	9	19	6	22

*indicates significant difference ($p < .05$), independent samples t-test used

[a] Asthma, Cerebral Palsy, Congenital Heart Disease, Dyslexia, Hydrocephalus

[b] Acquired Brain Injury, Asthma, Cancer, Crohn's Disease, Epilepsy, Heart Disease, Irritable Bowel Syndrome, High Blood Pressure, Rheumatoid Arthritis

The mean age of CWE (n = 48) included in the study was 9.9 years (SD ± 3.1), 60% of whom were female. On average CWE were taking 1.5 (SD ± 1.0) anti-epileptic medications (AEDs) and had a mean age of epilepsy diagnosis of 7.1 years (SD ± 5.4). The majority of CWE's seizures were well controlled with 40% reported to have had no seizure activity and 35% reported to have had less than 10 seizures in the previous three months. Eight per cent of CWE were reported to have had between 10 and 19

seizures and 17% had more than 20 seizures. The mean age of parents completing the questionnaires was 42.4 years (SD ± 5.5) who were 90% mothers. Seventy-one per cent of parents were living together, 59% were employed, and 72% had completed post-secondary school education.

The sample of questionnaires returned from teachers was generally representative of the overall sample, however CWE in the teacher sample were slightly younger in age (8.8 years ± 2.6) and so also had a younger age of diagnosis (5.7 years ± 2.9; $p < .05$). No significant differences were found between the two samples for any other sociodemographic or epilepsy related variables, including seizure frequency ($p > .05$). Additional clinical and sociodemographic characteristics are presented for both the overall sample of families and CWE, and the teacher subsample, in Table 3.

2.2 Ethical approval

Ethical approval for this study was granted by a University College Cork Social Research Ethics Committee (SREC; see Appendix 6). Eligible families ($n = 159$) were contacted and invited to participate by their Consultant Paediatric Neurologist via letter (see Appendix 7; including a study information sheet and consent form, see Appendix 8 and 9 respectively), or directly through epilepsy outpatient clinics if they presented to such during the recruitment phase. Consent forms were returned to the research team who then forwarded the study questionnaires as outlined below.

2.3 Study Measures

2.3.1 Sociodemographic factors

These were assessed through the bespoke study family demographic questionnaire which included information on: age; gender; country of birth; family composition;

marital status; employment status; parental education; and illnesses/diagnoses in parent or siblings (see Appendix 10). This information was transformed into dichotomous (yes/no) independent variables for analysis with the child outcome variables.

2.3.2 Disease / medical factors

These were identified from a bespoke medical history questionnaire which gathered information on seizure frequency, age at diagnosis, and anti-epileptic medication use (see Appendix 11).

i. Seizure frequency: This was recorded as the total number of seizures over the previous three months. As this was quite a heterogeneous variable, for the purpose of analysis it was categorised into four groups: i) no seizures, ii) <10 seizures, iii) 10-19 seizures, iv) <20 seizures.

ii. Anti-epileptic medication use: This was coded as a dichotomous variable (yes/no) for analytic purposes. Information was also collected on the number of anti-epileptic medications for each child.

2.3.3 Child outcome and predictor factors

i. Emotional and behavioural adjustment: The Strengths and Difficulties Questionnaire (SDQ) [48] is a brief 25-item screening questionnaire. It assesses children's mental health difficulties, psychological strengths, and the impact of emotional and behavioural difficulties, as measured via parent, teacher and child report forms. Both parent and teacher report forms were

utilised in the current research. Items are answered on a 3-point scale (0 = not true – 2 = certainly true), with higher scores reflecting greater difficulties, that can be categorised into '*Normal'*, *'Borderline'* and *'Abnormal'* bands. To reduce the number of categories for analysis in the current study, children scoring at or above *'Borderline'* thresholds were considered to be at risk of clinical levels of emotional and behavioural difficulties (in both parent and teacher reports) and classified as *'Clinical'*.

The *SDQ* has been standardised in large populations in the US and UK and has demonstrated good test-retest reliability and validity data [49,50]. It has been regularly used in other studies of CWE [51]and other chronic illnesses [52-54]. The current study found satisfactory internal consistency for the scale, reporting a Cronbach's alpha coefficient of 0.84 for the parent-reported total difficulties scale, and 0.68 for the teacher-reported total difficulties scale.

ii. Social competence: The competence items of the *Child Behaviour Checklist* (CBCL/6-18) [55] Parent Report Form (PRF) was used as a measure of child social adjustment to illness. The scale provides a *Total Competence* score (T-scores with a mean of 50 and a standard deviation of 10) which is comprised of three subscales (*Activities, Social and School*). Scores falling one standard deviation below the mean (<40) were considered to be in the clinical range. The competence scale has demonstrated good test-retest reliability and internal consistency (0.90) [55] and has been used extensively in the literature with epilepsy populations [56] and other chronic illnesses.

In addition to this, an *Adaptive Functioning* total score on the Teacher Report Form (TRF) parallels the *Total Competence* score on the PRF. This *Total Adaptive Functioning* score represents the amalgamation of four sub-scale scores pertaining to working hard, behaving appropriately, learning, and happiness. Furthermore, an academic performance competence sub-scale is also calculated. As with the *Total Competence* score, similar T-scores for the total adaptive functioning scale and the academic subscale were computed, with a score below 40 being considered as clinically significant. Similar to the competence scales, the adaptive functioning scales demonstrate excellent internal consistency and reliability (Cronbach's alpha coefficient = 0.90) [55].

iii. Quality of Life: The Pediatric Quality of Life Inventory - Epilepsy Module (PedsQL- Epilepsy Module) [57,58] is a 29-item, epilepsy specific, standardized generic assessment instrument that systematically assesses psychological, emotional and health-related quality of life domains in children and adolescents with epilepsy. It has five individual scales: *Impact, Cognitive, Sleep, Executive Function*, and *Mood/Behavior*, and items are scored on five point Likert scale (0 = none – 4 = almost always) with higher scores indicating higher quality of life. There is no total score for this measure. Modi and colleagues (2017) demonstrated excellent psychometric properties across the epilepsy spectrum in both clinical and research settings for the *Pediatric Quality of Life Inventory – Epilepsy Module*. They reported high Cronbach's alpha coefficients for each individual subscale, ranging from .77 to .94. This was echoed in the current study which found alpha coefficients of .89 (*Impact*), .96 (*Cognitive Functioning*), .77 (*Sleep*), .89 (*Executive*

Functioning), .87 (*Mood / Behaviour*). Whilst outcomes across all subscales were noted, the impact of epilepsy on quality of life was the outcome of greatest interest, and so the "*Impact*" subscale was employed as a dependent variable to represent quality of life for purposes of analysis in the current study.

2.3.4 Family outcome and predictor factors

i. Family functioning: The *Family Relations Index* (FRI) comprises of 27 items and is a well validated and recognised subscale of the *Family Environment Scale* (FES) [59]. The *FRI* looks at how family members relate to each other. Subscales included are *Cohesion*, defined as the '*family's degree of mutual commitment and the help and support family members provide for one another'*; *Conflict*, defined as the '*amount of openly expressed anger, aggression, and conflict among family members'*; and *Expressiveness,* defined as the '*extent to which family members are encouraged to act and express their feelings directly'* (p.2) [59]. The scale has demonstrated good internal reliability and consistency in the literature, with Cronbach's alpha coefficients ranging from .49 to .78 [60]. The current sample reported lower than expected alpha coefficients: .33 (*Cohesion)*, .49, *(Expressiveness),* .68 *(Conflict)*. However it has been noted previously by the scale authors [61] that lower alphas for this scale are adequate given that its aim is to address a construct reflecting a wide range of functioning.

ii. Family resilience: The *Family Resilience Assessment Scale* (FRAS) [62] is a 54-item scale that measures the construct of family resilience as conceptualised by Walsh [63] and specifically, processes that support a family's ability to cope

successfully with adversity. Items are rated on a 4-point scale (1 = strongly disagree – 4 = strongly agree), with higher scores representing higher levels of family resilience. A recent validation study provided support for this measure as a reliable and valid scale for assessing the construct of family resilience in paediatric epilepsy, reporting a Cronbach's alpha coefficient of 0.92 [64] albeit with young people with epilepsy (aged 13-16 years), rather than parents. A similarly good Cronbach's alpha coefficient of 0.91 was found for the total scale in the current study.

iii. Maternal worry: The *Maternal Worry Scale* (MWS) [65] is an 11-item self-report scale that measures parental worry in children with chronic illnesses. Items are rated on a four-point Likert scale from (1 = not at all – 4 = most of the time). Past research has indicated that parental worry can fluctuate independently of disease severity and affect behavioural adjustment independently [66]. It has demonstrated notably good internal consistency with a Cronbach's alpha coefficient of 0.94. The current study mirrored this, also producing an alpha of 0.94.

The full battery of psychometric questionnaires can be found in Appendix 12.

2.4 Design and statistical analysis

The study employed a cross-sectional survey style research design (see Appendix 13 for a more in-depth discussion of this methodology). However, historical factors (e.g. age of diagnosis, medications and treatments received, socioeconomic factors) were collected retrospectively for inclusion as predictor variables in statistical analysis.

All analyses were completed using SPSS version 25. On checking the statistical assumptions prior to data analyses, it was highlighted that the assumption of normality was violated for the total Impact score for the *PedsQL-Epilepsy*, and the *Cohesion* and *Conflict* scales of the *FRI*, as referenced by the Shapiro Wilk test ($p < .05$). The authors considered transformation of the data [67], however, after visual inspection of the data distribution, as presented via histogram, examining relative levels of skewness and kurtosis, and comparing the results of parametric and non-parametric tests, it was concluded that the data was satisfactory for parametric analyses, and so transformation was not required. No other violations of statistical assumptions were detected.

To determine whether the smaller teacher subset of data was representative of the entire sample, t-tests, one-way ANOVAs, or chi-square tests of independence were completed on sociodemographic variables (age, gender, age at diagnosis, seizure frequency, family composition, etc.) and psychological family outcomes (*FRI, FRAS, MWS*) and measures of psychosocial outcome (*SDQ, CBCL, PedsQL-Epilepsy*). Summary means and proportions in the clinically significant range were computed for emotional/behavioural adjustment (*SDQ*) and social competence/adaptive behaviour (*CBCL*, PRF and TRF) were noted. The clinical significance of these was highlighted by examining deviations from normative means, where available. Outcomes for CWE were also compared by level of seizure frequency to assess whether they were varying differentially by subgroup. No significant differences were found and so the sample was analysed as a whole for bivariate and multivariate analyses.

Given the relatively large number of predictive variables in relation to sample size, exploratory bivariate analyses were first carried out on all predictor and outcome

variables. This included parametric (Pearson's product moment correlations, t-tests) and non-parametric (Kendall's tau) analyses, which were completed according to the measurement of the data. An alpha level of .05 was used to determine significance and only significant factors were taken forward for multivariate analyses in order to assess the unique and combined power of the predictors in understanding child psychosocial outcomes.

Hierarchical multiple regression was used to assess the predictive ability of variables which emerged as significant following bivariate analyses on child psychosocial outcomes. Three individual higher order multiple regressions were conducted on the parent-reported child outcomes (emotional/behavioural adjustment – *SDQ Total Difficulties*, total social competence – *CBCL Total Competence*, and quality of life – *PedsQL – Epilepsy Impact* subscale). Similarly, two multiple linear regressions were carried out to assess the predictive ability of variables which were significant at the bivariate level and teacher-reported child outcomes (emotional/behavioural adjustment – *SDQ Total Difficulties*, total academic performance – *CBCL Total Academic*). All statistical assumptions were met with satisfaction for multivariate analyses.

3. Results

3.1 Child and family outcomes

Child outcomes, as reported by parents and teachers, are summarised in Table 4 and compared to population based normative data (where available). No significant differences were found between the overall sample and the teacher subsample with regard to parent-reported psychosocial outcomes and psychological family factors. In

terms of reported outcomes for child emotional and behavioural adjustment (*SDQ*), parents and teachers were most concerned regarding perceived *Emotional Difficulties* for CWE. The magnitude in difference of these scores to that of the normative population was found to be considerably large (d = -1.4). Parents rated perceived *Peer Difficulties* as the next most concerning with 48% of children reported to be in the clinical range, along with *Conduct* difficulties (44% in clinical range). *Hyperactivity/Inattention* levels were noted by parents to be least prevalent, which was not found to be unduly different to the general population (d = -.2). Overall teachers reported difficulties to a lesser degree, however, were in agreement that *Emotional Difficulties* were most prevalent. Interestingly, teachers reported difficulties with peer relations and conduct as the least prevalent and instead attributed more weight to difficulties with *Hyperactivity/Inattention*. Effect size deviations between parent and teacher-reported outcomes ranged from -.2 – 1.0 (Cohen's d) [68]. A full comparative summary of parent and teacher scores can be found in supplementary Table 2 (Appendix 14).

In total parents rated 65% of children to be in the clinical range in relation to overall social competence scores (*CBCL*), which also represented a large difference in effect to that of the normative population (d = -1.2). Furthermore, difficulties with *School* functioning were noted to be most prevalent in the sample with 73% of children reported to have difficulties reaching clinical level, followed by involvement and skill level in *Activities* and *Social* functioning. Paralleling parent-reported competence outcomes were teacher-reported academic and adaptive functioning skills. Again, teacher-reported outcomes were to a lesser extent, however reflected similar patterns. For example, academic difficulties were most prevalent as noted by scores for *Total*

Academic Performance (60% in the clinical range) and *Learning* (37% in the clinical range).

With regard to outcomes in quality of life (*PedsQL-Epilepsy*), children in the current sample were found to be functioning similarly to other CWE across all scales (effect size deviations were small and ranged from d = -.01 to -.4) with the exception of the *Mood / Behaviour* subscale, for which a medium effect size deviation was noted (d= -.6). No comparative data was available with a sample of children from the general population as this measure is designed solely for use with CWE.

Table 4: Summary of mean parent-reported outcomes scores, including the percentage of children in the clinical range, and corresponding normative population scores

Scale	Mean ± SD	% in clinical range (where appropriate)	Normative population Mean ± SD	Cohen's *d*
Parent report (*n* = 48)				
SDQ				
Prosocial	7.6 ± 2.4	25	8.6 ± 1.6[a]	-.6**
Peer difficulties	2.5 ± 1.9	48	1.5 ± 1.7[a]	.6**
Hyperactivity/inattention	4.1 ± 2.5	23	3.5 ± 2.6[a]	.2*
Conduct	2.5 ± 2.2	44	1.6 ± 1.7[a]	.3*
Emotional difficulties	4.7 ± 2.8	65	1.9 ± 2.0[a]	1.4***
Total difficulties	13.9 ± 6.7	50	8.4 ± 5.8[a]	.9***
CBCL - PRF				
Activities	40.6 ± 9.2	50	50 ± 10[b]	-.9***
Social	44 ± 9.7	38	50 ± 10[b]	-.6**
School	35 ± 9.3	73	50 ± 10[b]	-1.5***
Total Competence	37.6 ± 10.8	65	50 ± 10[b]	-1.2***
PedsQL - Epilepsy				
Impact	66.3 ± 23.1	-	73.3 ± 24.4[c]	-.3*
Cognitive functioning	48.8 ± 34.1	-	57.1 ± 31.0[c]	-.3*
Sleep	55.8 ± 24.9	-	56.1 ± 26.3[c]	-.01*
Executive functioning	43.5 ± 28.2	-	56.1 ± 28.1[c]	-.4*
Mood / Behaviour	50 ± 23.3	-	64.3 ± 21.5[c]	-.6**
Teacher report (*n* = 27)				
SDQ				
Prosocial	8.1 ± 2.11	11	7.2 ± 2.4[a]	.2*
Peer difficulties	.9 ± 1.37	7	1.4 ± 1.8[a]	-.9***
Hyperactivity/inattention	3.6 ± 2.19	18	2.9 ± 2.8[a]	-.2*
Conduct	.5 ± 1.12	7	0.9 ± 1.6[a]	-1.0***
Emotional difficulties	3.2 ± 2.59	26	1.4 ± 1.9[a]	-.5**
Total difficulties	8.3 ± 5.41	30	6.6 ± 6.0[a]	-.9***
CBCL – TRF				
Total Academic Performance	43.3 ± 6.90	60	50 ± 10[b]	-.7**
Working hard	45.9 ± 7.70	30	50 ± 10[b]	-.5*
Behaviour	49.2 ± 8.09	7	50 ± 10[b]	-.1*
Learning	43.7 ± 7.63	37	50 ± 10[b]	-.7**
Happy	48.9 ± 7.79	7	50 ± 10[b]	-.1*
Total Adaptive Functioning	45.1 ± 7.81	23	50 ± 10[b]	-.5**

SDQ = Strengths and Difficulties Questionnaire, CBCL = Child Behaviour Checklist, PRF = Parent Report From, PedsQL – Epilepsy = Pediatric Quality of Life Inventory – Epilepsy Module, TRF = Teacher Report Form

[a] Meltzer, Gatward, Goodman, & Ford, 2000 [50]

[b] Scores for both groups represent T-scores which are standardised with a mean of 50 and a standard deviation of 10

[c] Modi et al., 2017 (sample is from a population of children with epilepsy – no normative data available from the general population) [58]

*small effect size, **medium effect size, ***large effect size

Table 5 reports outcomes for psychological family factors, including means and corresponding normative population data (where available). These results suggested that families of CWE in the current study reported total family resilience scores (FRAS) slightly higher than the normative data reported for the general population. However, the magnitude of this difference was noted to be small (*d* = .4). Furthermore, family functioning results as measured by the *FRI* subscales of *Cohesion,*

Expressiveness and *Conflict*, were also found to be relatively similar to general population group norms, with effect size differences noted to be small, ranging from -.1 to .3 (Cohen's *d*). Parental worry scores in the current sample were found to be at the higher end of the scale, however no normative population data was available for comparison.

Table 5: Summary of mean parent-reported family outcomes scores, including corresponding normative population scores

Scale (n = 48)	Parent report Mean ± SD	Normative population Mean ± SD	Cohen's *d*
FRAS			
FCPS	83.6 ± 8.1	78.6 ± 11.1[a]	.5**
USER	23.7 ± 4.4	23.3 ± 3.7[a]	.1*
MPO	19 ± 2.7	19.5 ± 2.5[a]	-.2*
FC	18.2 ± 2.2	15.1 ± 1.7[a]	1.7***
FS	9.3 ± 2.6	9.9 ± 1.4[a]	-.3*
AMMA	10.2 ± 1.4	9.9 ± 1.4[a]	.2*
Total scale	163.9 ± 14.3	157.5 ± 17.4[a]	.4*
FRI			
Cohesion	7.2 ± 1.4	6.73 ± 1.5[b]	.3*
Expressiveness	5 ± 2.1	5.54 ± 1.6[b]	-.3*
Conflict	2.9 ± 2.1	3.18 ± 1.9[b]	-.1*
MWS			
Total scale	27.1 ± 8.3	N/A	-

FRAS = Family Resilience Assessment Scale, FCPS = Family Communication and Problem Solving, USER = Utilising Social and Economic Resources, MPO = Maintaining a Positive Outlook, FC = Family Connectedness, FS = Family Spirituality, AMMA = Ability to Make Meaning of Adversity, FRI = Family Relations Index, MWS = Maternal Worry Scale, N/A = not available

[a] Sixeby, 2005 [62]

[b] Moos & Moos, 2009 [69]

*small effect size, **medium effect size, ***large effect size

3.2 Exploratory bivariate analyses

Exploratory bivariate analyses were carried out on all predictor and parent-reported child medical and psychosocial outcome variables, a summary of which is presented in Table 6. Statistically significant associations were found between a number of predictor and outcome factors ($p < .05$). Child emotional/behavioural adjustment produced significant negative associations with respondent age, education, employment, lone parent, and child total social competence and a positive association with parental worry. Relationships between parent-reported child emotional/behavioural adjustment and the presence of an illness or diagnosis in either parent, the number of children in the family, child gender, age, age at diagnosis,

seizure frequency (reported for the previous three months), family functioning and family resilience did not emerge as statistically significant. Seizure frequency and parental worry were found to be statistically significantly negatively associated with child social competence. A statistically significant positive association emerged between child quality of life and lone parent, while statistically significant negative associations were found for seizure frequency and parental worry and child quality of life. Associations with all other structural and psychological family factors and outcomes did not reach statistical significance. Full details of the strength of the associations can be found in Table 6.

Table 6: Summary of exploratory associations of predictor and outcome variables using parent-reported outcome measures

	SDQ Total Difficulties	CBCL Total Competence	PedsQL Epilepsy Impact
Respondent age	-.29[a]*	-.02[a]	-.07[a]
Respondent education	2.7[a]**	-1.19[a]	-.45[a]
Respondent employment	-2.5[b]**	1.25[b]	.49[b]
Lone parent	-2.6[b]*	1.14[b]	2.41[b]*
Illness/diagnosis in parent	-.100[b]	-1.19[b]	0.12[b]
Number of children in family	-.01[a]	-.15[a]	-.19[a]
Gender of CWE	.22[b]	1.51[b]	1.14[b]
Age of CWE	-.08[a]	.12[a]	-.24[a]
Age at dx	-.24[a]	.17[a]	-.11[a]
No. of meds	-.03[a]	-.18[a]	-.19[a]
Sz Frequency			
3M	.05[c]	-.22[c]*	-.31[c]**
CBCL			
Total Competence	-.46[a]**	-	.22[a]
FRI			
Cohesion	-.08[a]	.00[a]	.02[a]
Expressiveness	-.09[a]	.14[a]	-.08[a]
Conflict	.13[a]	-.21[a]	-.12[a]
FRAS			
Total Scale	.21[a]	.18[a]	-.06[a]
MWS			
Total score	.31[a]*	-.36[a]*	-.48[a]**

SDQ = Strength and Difficulties Questionnaire, CBCL = Child Behaviour Checklist, PedsQL – Epilepsy = Pediatric Quality of Life Inventory – Epilepsy Module, CWE = Child with Epilepsy, Sz = seizure, 3M = 3 months, FRI = Family Relations Index, FRAS = Family Resilience Assessment Scale, MWS = Maternal Worry Scale

[a] = Pearson's r

[b] = t test

[c] = Kendall's tau

* $p < 0.05$

**$p < 0.01$

Similar exploratory analyses were also completed for teacher-reported outcomes. Significant correlations ($p < .05$) were found between teacher-reported child emotional/behavioural adjustment and seizure frequency, and social competence scores. No significant associations were found between child total *Adaptive Functioning* and any of the predictor variables, while child total *Academic Performance* scores were found only to be significantly correlated with seizure frequency. Moreover, no significant relationships were found between psychological family factors (family functioning, family resilience, parental worry) and teacher-reported child outcomes. The full results of these exploratory correlations between predictor and teacher-reported psychosocial outcomes are presented in Table 7.

Table 7: Summary of associations of predictor and outcome variables using teacher-reported outcome measures

	SDQ Total Difficulties	CBCL Total Adaptive Functioning	CBCL Total Academic
Respondent age	.24	.06[a]	.25[a]
Respondent education	1.22[b]	-.53[b]	-.29[b]
Respondent employment	.48[b]	-.91[b]	-1.21[b]
Lone parent	1.85[b]	.00[b]	-.49[b]
Illness/diagnosis in parent	.89[b]	-.88[b]	-.88[b]
Number of children in family	.27[a]	-.07[a]	-.31
Gender of CWE	.07[b]	.87[b]	.28[b]
Age of CWE	-.03[a]	-.02[a]	.16
Age at dx	-.24[a]	.22[a]	.38
No. of meds	-.08[a]	-.00[a]	-.22
Sz Frequency			
3M	.33[c]*	-.26[c]	-.32[c]*
CBCL			
Total Competence	-.43[a]*	.32[a]	.21[a]
FRI			
Cohesion	.25[a]	.07[a]	.07[a]
Expressiveness	-.13[a]	.10[a]	.10[a]
Conflict	.07[a]	-.08[a]	.33[a]
FRAS			
Total Scale	.-18[a]	.26[a]	-.05[a]
MWS			
Total score	.17	-.15	-.12[a]

SDQ = Strengths and Difficulties Questionnaire, CBCL = Child Behaviour Checklist, CWE = Child with Epilepsy, Sz = seizure, 3M = 3 months, FRI = Family Relations Index, FRAS = Family Resilience Assessment Scale, MWS = Maternal Worry Scale

[a] = Pearson's r

[b] = t test

[c] = Kendall's tau

* $p < 0.05$

3.3 Multivariate analyses

Only those factors that demonstrated significance in the exploratory analyses were entered into the multiple regressions. Relevant structural family factors and/or medical factors were entered into the first block, with psychological family factors and child competence factors entered in the second block. This was to enable the assessment of the unique and combined contributions of psychological factors to child outcomes.

3.3.1 Factors predicting child emotional and behavioural adjustment – parent and teacher-reported outcomes

Results of hierarchical multiple regression illustrated that respondent age, education, employment and lone parent significantly explained 36.7% of the variance in parent-reported child emotional/behavioural adjustment. However, when parental worry and child total competence were added to the model, the total variance explained increased to 48.6%, indicating that psychological factors explain an additional 11.8% of the total variance in parent rated child emotional/behavioural adjustment (R^2 change = .118, F change [2, 39] = 4.487, p = 0.018) . The final model was significant and illustrated that parent education and child competence significantly negatively predicted child emotional/behavioural adjustment, with both variables contributing similarly. Full details of the strength and significance of relationships can be found in Table 8.

Table 8: Results of hierarchical multiple regression predicting Strengths and Difficulties Questionnaire Total Difficulties score (parent report, *n* = 48)

	SDQ Total Difficulties – Parent report					
	Model 1			Model 2		
	β	*t*	*p*	β	*t*	*p*
Respondent age	-.179	-1.400	.169	-.198	-1.667	.104
Respondent education	**-.323**	**-2.564**	**.014**	**-.269**	**-2.276**	**.028**
Respondent employment	**.251**	**1.981**	**.054**	.201	1.697	.098
Lone parent	**.292**	**2.282**	**.028**	.224	1.856	.071
MWS total score				.118	.953	.347
CBCL Total Competence (parent)				**-.301**	**-2.363**	**.023**

SDQ = Strengths and Difficulties Questionnaire, CBCL = Child Behaviour Checklist, MWS = Maternal Worry Scale
Model 1 = Structural family factors
Model 2 = Structural family factors + psychological family factors
Bold values represent significant results
Note: R^2 for model one is .367, Adjusted R^2 = .306, p = .001 and for model 2 is .486, R^2 = .407 p < .0001

In terms of teacher-reported adjustment, just two factors (seizure frequency and total social competence) were entered into a multiple linear regression model, with the overall model reaching significance, explaining 29.5% of the total variance in outcome (see Table 9). Although seizure frequency was found to be approaching significance (p = .059), child total social competence was the only variable to make a statistically significant and unique contribution to teacher-reported emotional/behavioural adjustment. This result is similar to the parent-reported model in Table 8, which also found that child social competence was the most significant predictor of adjustment difficulties.

Table 9: Results of multiple linear regression predicting Strengths and Difficulties Questionnaire Total Difficulties score (teacher report, *n* = 27)

	SDQ Total Difficulties – Teacher report		
	β	*t*	*p*
Sz in previous 3M	.344	1.978	.059
CBCL Total Competence (parent)	**-.366**	**-2.107**	**.046**

CBCL = Child Behaviour Checklist, Sz = seizure, 3M = 3 months
Bold values represent significant results
Note: R^2 = .295, Adjusted R^2 = .237, p = .015

3.3.2 Factors predicting child social competence – parent and teacher-reported outcomes

In predicting child overall social competence, the result of the first hierarchical model, which contained medical variables alone, was not significant (p = .192). However, the

addition of parental worry in the second block, rendered the overall model significant, suggesting that, together, the presence of seizures and parental worry explained 13.6% of the variance in child social competence. Yet, only parental worry made a significant contribution to total social competence. This finding suggests that parental worry predicts the total social competence (*Activities, School* and *Social* competence) of CWE over and above that of seizure frequency. Specific details of the regression model and strength of associations can be found in Table 10.

Table 10: Results of hierarchical multiple regression predicting Child Behaviour Checklist Total Competence score (parent report, *n* = 48)

	CBCL Total Competence – Parent report					
	Model 1			Model 2		
	β	*t*	*p*	β	*t*	*p*
Sz in previous 3M	-.194	-1.326	.192	-.095	-.643	.523
MWS total score				**-.328**	**-2.234**	**.031**

CBCL = Child Behaviour Checklist, Sz = seizure, 3M = 3 months, MWS = Maternal Worry Scale
Model 1 = Medical factors
Model 2 = Medical factors + psychological family factors
Bold values represent significant results
Note: R^2 for model one is .038, Adjusted R^2 = .016, p = .192 and for model 2 R^2 is .136, Adjusted R^2 = .096, p = 0.040

A linear regression (see Table 11) was conducted to determine whether the association between seizure frequency and child academic performance (as rated by teachers) remained under more robust analysis. The overall model was significant and suggested that seizure frequency explained 16.8% of the variance in outcome. This suggests that the frequency of seizures experienced in CWE is important to consider when thinking about academic competence.

Table 11: Results of linear regression predicting Child Behaviour Checklist Total Academic Performance (teacher report, *n* = 27)

	CBCL Total Academic Performance – Teacher report		
	β	*t*	*p*
Sz in previous 3M	**-.410**	**-2.158**	**.042**

CBCL = Child Behaviour Checklist, Sz = seizure, 3M = 3 months
Bold values represent significant results
Note: R^2 = .168, Adjusted R^2 = .132, p = .042

3.3.3 Factors predicting child quality of life

All variables, which included structural family factors (lone parent), medical factors (presence of seizures in the previous three months) and psychological family factors (parental worry), significantly predicted child quality of life, with parental worry emerging as the most significant predictor. Furthermore, parental worry contributed an additional 10.8% to the variance of the total model (R^2 change = .108, F [1,44] = 7.423, p = .009), which suggests that parental worry is uniquely related to child quality of life beyond that of structural family factors and medical factors in this sample, see Table 12 for full details of the model.

Table 12: Results of hierarchical multiple regression predicting Pediatric Quality of Life Inventory – Epilepsy Module Impact score (parent report, n = 48)

	PedsQL – Epilepsy Impact – Parent report					
	Model 1			Model 2		
	β	*t*	*p*	β	*t*	*p*
Lone parent	**-.351**	**-2.712**	**.009**	**-.285**	**-2.307**	**.026**
Sz in previous 3M	**-.370**	**-2.860**	**.006**	**-.261**	**-2.044**	**.047**
MWS total score				**-.352**	**-2.724**	**.009**

PedsQL – Epilepsy = Pediatric Quality of Life Inventory – Epilepsy Module, Sz = seizure, 3M = 3 months, MWS = Maternal Worry Scale
Model 1 = Structural family factors + medical factors
Model 2 = Structural family factors + medical factors + psychological family factors
Bold values represent significant results
Note: R^2 for model one is .249, Adjusted R^2 = .215, p = .002 and for model 2 R^2 is .357, Adjusted R^2 = .313, p < .001

4. Discussion

4.1 Profile of Outcomes

Children with epilepsy are suggested to have four times the risk of developing psychosocial difficulties than typical peers [9]. The profile of psychosocial difficulties in CWE in the current study supports this, portraying more pronounced difficulties in comparison to population-based norms. However, the current sample also present with difficulties that are higher in frequency to those detailed within the epilepsy literature for emotional/behavioural adjustment (50% in the current sample versus 37% in a UK nationwide sample of CWE [70], and particularly for social competencies. The current

sample noted that 65% of CWE were in the clinical range for difficulties with social competence, in comparison to 30-33%[71], 31% [72], and 28% [73] in previous studies.

Looking closer at emotional/behavioural adjustment, emotional difficulties were noted to be the most prevalent difficulties in the current sample by parents and teachers. This is consistent with previous literature which outlines a higher incidence of emotional rather than behavioural problems in childhood epilepsy [2,3,5,74]. Difficulties with hyperactivity/inattention were not an issue for children in this sample as rated by their parents. This is in contrast to previous findings which report difficulties of this type as most prevalent [15,31]. However, this may be due to the study's exclusion criteria which excluded children who had a comorbid diagnosis of ADHD. Interestingly, the subsample of teacher-reported outcomes did emphasise hyperactivity/inattention difficulties as the second most prevalent concern. It is possible that such difficulties may be more noticeable to teachers in that they are likely to cause more disruption to the classroom environment and so need more direct management time from teachers.

Peer difficulties were noted to be quite prevalent in this sample of children and young people as noted by their parents, with nearly half reportedly experiencing difficulties in the clinical range. This was consistent with previous findings, whereby 39% of the sample were experiencing similar difficulties, second only to difficulties of hyperactivity and inattention [15]. These results were not seen however in teacher reports (7%) in the current study. Previous literature has suggested that, peer difficulties, particularly when they relate to social skills deficits, can often go unnoticed by teachers as they do not directly cause behavioural difficulties, such as

acting out [72]. It is possible that this could account for the discrepancy in scores between parent and teacher ratings of peer difficulties. In a similar vein to the previous point regarding difficulties of hyperactivity/inattention requiring more time from teachers, children with such peer difficulties that are not as directly observable could be 'flying under the radar' in busy classrooms.

Difficulties with overall social competence appear to be of most concern to parents in the current sample (65% in the clinical range versus 50% for adjustment difficulties). As noted, previous studies measuring social competence (also using the *CBCL* competence scales) [71-73] in CWE did not report the same level of deficits as the current study. A meta-analysis [75] of social competence in children with chronic illnesses reported that children with neurological disorders had the lowest levels of social competence of all included illnesses (e.g. obesity, cancer, diabetes, asthma). For the current parent and teacher samples, *School* related difficulties appeared to be the most prevalent in the competence subscales. This finding should be considered regarding the attainments of CWE and thinking about their need for individualised support plans to enable them to reach their full potential in achieving academic success. It is especially important given that previous research has indicated that adults who have experienced childhood onset chronic illnesses and who have had poorer outcomes and experiences with schooling, can have higher rates of unemployment [76,77] and psychological difficulties in adulthood [78].

It is well documented in the literature that CWE experience poorer quality of life than their healthy peers [16,35,79]. Children in the current sample were reported to have similar scores in domains of quality of life to children in other epilepsy samples which

used the PedsQL – Epilepsy [58]. Even though no data was available to compare this with population norms using the PedsQL-Epilepsy, Huang and colleagues [80] generated clinical cut off scores by age group for the generic form of the Pediatric Quality of Life Inventory (PedsQL – 4.0; [81]) and suggested that scores below 79 (< 8 years old) and below 76 (> 8 years old) were clinically meaningful for children with a moderate chronic health condition. Utilising such cut off frameworks with the current sample would suggest that the CWE are experiencing a clinically meaningful impact on quality of life across all domains, albeit still less pronounced (as noted by effect size deviations) than difficulties with emotional/behavioural adjustment or social competence.

In terms of family-based outcomes, the current sample reported family functioning and family resilience levels relative to that of population-based norms. Family resilience levels, as measured by the FRAS, were also similar to that of a sample of families of youth with epilepsy in Singapore. However, notably, it was the young people themselves, and not the parents who reported for this sample. Nevertheless, the current sample appears to have been functioning well as families, and in general were quite highly resourced (e.g. almost two-thirds of respondents in employment and nearly three-quarters of the sample having completed post-secondary school education). These positive findings in relation to typical levels of family functioning and resilience may reflect what is referred to as 'post-traumatic growth' – the process of attributing meaning to traumatic life events (such as receiving a diagnosis of chronic illness) perceiving some benefit from it, and incurring some positive life developments [82]. It has been noted in the literature with other chronic illnesses, that families who have learned through adversity experience better levels of family functioning

(communication and satisfaction with family relationships) which are likely to reflect post-traumatic growth, and often can promote better adjustment [83,84].

Lastly, although parental worry scores in the current sample could not be compared to population based norms, or directly with other epilepsy populations, they were found to be higher than that of mothers of children with diabetes, asthma, sickle cell disease and cystic fibrosis [85] and substantially higher than children with Congenital Heart Disease [66] and children with newly diagnosed cancer [86], all of which utilised the *MWS*. This suggests that parents in this sample were notably more worried than parents of children with other chronic health conditions. This finding could perhaps be due to the unpredictable and somewhat sporadic nature of seizures, as described earlier. Regardless, it is an important area to be targeted at intervention level given the previous associations in the literature concerning increased parental worry about epilepsy and poorer child outcomes, including lower quality of life [35,87], lower social competence [88] and lower adaptive functioning [89] as well as increased emotional/behavioural difficulties [34].

4.2 Psychological family factors predicting child psychosocial outcomes

Results of the exploratory analyses indicated that parental worry was the only factor to be associated with all parent rated child outcome measures (emotional/behavioural adjustment, social competence and quality of life), while seizure frequency was associated with two out of three outcomes (social competence and quality of life). On the other hand, exploratory analyses for teacher-reported outcomes highlighted a different narrative, finding that child social competence and seizure frequency were the only variables to be significantly associated with emotional/behavioural

adjustment and academic performance. These exploratory results suggest that when independent respondents for child outcomes were utilised, seizure factors, and individual child factors, were most consistently associated with child outcome. These findings may be the result of fewer reported difficulties in the teacher-reported sample in general compared to parent reports. It may also be that differential results reflect particular factors specific to the school environment that may perhaps contain children's overt expression of difficulties. For example, school settings are likely to more consistently enforce strict rules and boundaries for acceptable behaviour than a home environment where parents are already worried/anxious. Moreover, it may be that parents who are worried about their CWE are more sensitive to noticing any potential difficulty in their child, and perhaps have a tendency to over-report their level of difficulty.

Regression analysis was used to identify predictors of child psychosocial outcomes from psychological family factors. Factors, which showed the strongest relative associations with parent-reported emotional/behavioural adjustment, were parental education and child social competence, when controlling for parental worry, respondent age, employment and lone parent families, which has been replicated previously[25,71,72]. The entire model as a whole explained almost half of the variance in child adjustment, with child social competence producing the strongest unique contribution. Similarly, child social competence was found to be the most important unique predictor of adjustment, over and above that of seizure frequency in teacher-reported outcomes also. This suggests that CWE's capacity to develop appropriate social skills, engage in activities and relationships with their peers and perform academically, negatively impacts their emotional/behavioural functioning

across settings. This is not surprising given the range of deficits in social competency noted in this sample. As a result, it is understandable that such children would struggle emotionally as this is undoubtedly a stressful experience for them. Importantly, the mechanisms by which this relationship occurs are in fact more critical than the severity of their epilepsy, their family environment and sociodemographic background.

Parental worry was the only unique predictor of social competence. This suggests that although parental worry was more strongly related to child social competence than medical factors in this study, there are certainly other factors responsible for determining the low scores found in this population. It has been suggested that decreased social competence in CWE may be the result of overprotective parenting styles, as CWE may be prevented from experiencing typical opportunities for social engagement for fear of seizure occurrence [47]. Such processes may also shed light on the findings in relation to child quality of life, where again, parental worry emerged as the most significant predictor (followed by lone parent families and seizure frequency). Although this was not measured directly, it is possible that parental worry may have enforced more overprotective parenting in these families which thus contributed to lower social competence and decreased quality of life. It may also be that parents who are more worried by nature do not themselves model effective social skills for their children to learn. Future research may wish to account for such variables as mediators of outcome, particularly given the high prevalence of social competence deficits in this population.

Teacher-reported outcomes for child competence fell under the categories of academic performance and adaptive functioning. As no factors were associated with adaptive

functioning, it was not possible to investigate this further, however, results may reflect teachers not perceiving a high rate of difficulties with this small sample. With regard to academic performance, seizure frequency was the only variable to show significant association, which also held true under more vigorous regression analysis. Previous studies investigating the impact of epilepsy variables on academic competencies have been mixed [32,33]. Furthermore, to limit the number of predictors and outcomes in the current study due to the small sample size, the *School* subscale of the *CBCL-PRF* was not examined individually, but rather under the umbrella of *Total Competence,* and also as more psychosocial factors, rather than academic factors, were outcomes of primary interest for the authors. Therefore, as such, a direct comparison for parent-reported outcomes is not reported.

It is surprising that the family functioning subscales (cohesion, expressiveness and conflict) or family resilience did not portray any significant relationships with child outcomes in this study, given the wealth of literature that documents such associations (Personal Correspondence). One reason for this may be the lower than expected levels of internal consistency in the *FRI* found for this sample (α = .33 - *Cohesion*, .49 – *Expressiveness,* and .68 - *Conflict*). It is possible that this scale may not accurately capture the process of family functioning for this particular population, particularly for the *Cohesion* subscale. Further research with larger Irish epilepsy populations may clarify this issue.

4.3 Limitations

There are a few important limitations pertaining to the current study that are necessary to mention. The study utilised an exploratory, cross-sectional design and so causality

between variables cannot be definitively determined. Thus, it must also be considered that relationships between family factors and outcomes for CWE in the current study are likely bi-directional in nature. However, the fact that social competence emerged as the strongest predictor of child adjustment difficulties for both parents and teachers suggests that this is a valid and reliable finding. Furthermore, results of prospective research which has considered the relationships between such variables has suggested that family factors continue to contribute to child outcomes over time. For example, increased parent/family stress, and parental fears and concerns about epilepsy have been found to consistently predict child quality of life more so than epilepsy and demographic factors when measured over a two year period [35]. Other prospective research has also advocated for parental worry as the strongest predictor of emotional/behavioural adjustment over time [34], and parental depressive symptoms as the strongest predictor of quality of life over time, again over and above the contribution of epilepsy factors [1]. Nonetheless, further prospective research will be important in obtaining a more thorough picture of this populations strengths and needs over time in an Irish context, for which the current paper provides a crucial starting point for such longitudinal work.

The study recruited from two of the major teaching hospitals in Munster as well as advertising on the Epilepsy Ireland webpage so as to ensure as representative a sample as possible given the scope of the project. Unfortunately, the resulting sample size was still small. Further, the study included multiple seizure types, as the small nature of the sample did not permit specification of particular types or severities, which perhaps could have impacted child outcomes differentially. That said, the current sample was found to be representative of the wider sample of families that chose not to participate

in the study with regard to both age and gender of CWE and so is promising with regard to the generalisability of findings to the wider population of families of CWE.

The Maternal Worry Scale (MWS) was used in the current study as a measure of parental worry. Although the majority of the reporting sample were mothers (94%), 6% were fathers and reported on the *MWS*. As the original validation study noted they did not have sufficient resources to calculate psychometric properties for fathers, no further data is available. However, the authors did clearly state that the scale was intended to be applicable to both parents [65]. Further, the scale itself does not specify gender and is focused on the chronically ill child, which indicates its application for both genders. Nevertheless, the lack of explicit validation/standardisation with fathers must be acknowledged.

The use of parental report for child difficulties in the absence of child self-report has been criticised in some previous literature. However research by Ferro and colleagues [1] noted similar reported outcomes from parents and children over a 28 – month period. This suggests that using parents as 'proxy-informants' for their children is valid and reliable. This finding, along with the use of teacher reports in the current study confer a strength in providing non-biased, independent ratings of child difficulties in conjunction with parental report.

4.4 Clinical implications

Despite the limitations outlined, the study provides a valuable insight into the profile of a sample of CWE in Ireland. To the best of our knowledge this is the first such study in an Irish context and will serve as a baseline for future research.

Significant associations between child outcomes and a number of factors were noted. Specifically, parental worry about epilepsy was noted to have a significant association with all outcomes, which remained significant at multivariate level with child quality of life and social competence. Parental education, lone parent and seizure frequency also remained significant at multivariate level. Results from the teacher-reported sample emphasised the importance of clinical factors in child outcomes, while both parents and teachers were in agreement that child social competence is an area regarding support to improve adjustment outcomes. These findings are important to note when considering interventions aimed at improving overall child adjustment and quality of life in epilepsy. They emphasise the need for a multi-layered approach which addresses psychological factors, such as parental worry, which is more amenable to change, but also incorporates psychoeducation, for example from the medical field, in upskilling parents in their knowledge of epilepsy. It also emphasises a role for teachers in promoting and modelling healthy social engagement in activities and relationships between CWE and their peers.

Moreover, as families in the current study appear to be well functioning and have high levels of resilience, future interventions should aim to bolster these positive factors and build on family's strengths. Research from the wider resilience and illness field suggests that implementing interventions that aim to support family strengths and build on these resources can also facilitate coping in families who are struggling to adapt to illness [90]. A systematic review and meta-analysis [28] found that psychological therapies with parents of children with chronic illnesses are beneficial in reducing parental distress and improving parenting behaviours. In particular,

building parents' problem-solving skills was noted to reduce parent distress long-term. A recent pilot study [91] which evaluated the feasibility, acceptability and outcome of a new psychosocial intervention (adapted from an established intervention for children with other chronic illnesses [92]) to strengthen family functioning in parents of CWE, found promising results. Following the one-day parent workshop and individual follow-up sessions, parents noted a decrease in worry about their CWE, as well as increases in family functioning measures. Encouraging positive shifts on child adjustment and quality of life outcomes were also noted. Although more research is needed in evaluating the effectiveness of such interventions for CWE over time, these results certainly indicate a positive movement toward improving outcomes for CWE and their families.

Furthermore, it has also been suggested that the provision of family centred care improves outcomes for children in paediatric neurology settings, independently of illness severity [93]. In keeping with this and the findings from the current study, incorporating brief screening measures for psychosocial outcomes and parental worry levels into routine outpatient neurology appointments would facilitate a more individualised approach to family care in hospitals and in doing so help front-line clinicians to identify families in need of further support.

4.5 Conclusion

The current study provides an insight into the profile of children and young people with epilepsy attending a paediatric outpatient service in Ireland. In doing so it highlighted that this sample appeared to have increased difficulties in relation to emotional and behavioural adjustment and quality of life than the typical population

but are functioning similar to other epilepsy populations. With regard to levels of social competence, children and young people in the current sample appear to be functioning at a lower level than both their peers with epilepsy and the general population – which identifies a specific area of need to be supported in interventions.

Positively, families in the current sample demonstrate high levels of functioning and resilience and so appear to be doing well in the face of adversity. However, high levels of parental worry were certainly evident in relation to their child's illness, which should be a target for future interventions in the field given the relationship noted at both bivariate and multivariate levels of analysis. Both parents and teacher-reported results indicate that child social competence is crucially important for healthy adjustment in CWE. Promoting positive social engagement and appropriate modelling of social skills across home and school settings is likely to have a positive impact here. The subsample of teacher data also emphasised the importance of seizure factors, indicating that a multi-layered approach is necessary to ensure that all levels of need are targeted at intervention. This would include psychoeducation from the medical and psychological fields, as well as psychological support for parents and families.

In all, and taking into account the limitations noted, the current study marks a positive step in supporting families of CWE in Ireland and paves the way for further research in the area aimed at supporting families and healthy psychosocial development of children and young people.

Disclosure of conflicts of interest

None of the authors had conflicts of interest to disclose.

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Appendices

Appendix 6: Letter of Ethical Approval

04/12/2017

Dear Emma,

Thank you for presenting your work to the D.Clin. Research Ethics committee and I am sorry for the rather tardy response.

The committee is happy to approve your research study but we would like to ask that you consider the points below and make some minor amendments to your protocol. You do not need to resubmit these changes for approval but we would ask that you do send us an electronic copy of your protocol with any such changes in place. Good luck with your research.

Sean Hammond

Please consider:

- The information sheet should clearly state that the research is independent from treatment and emphasize that "it will not affect access to or quality of" treatment received.
- Invitation letter is a little heavy handed and you may need to change the approach slightly and not give out all the questions at the start, but obtain consent before sending the full questionnaire.
- Consider the need to be more sensitive around wording in terms of vulnerable families.
- Please ensure that families don't feel coerced to participate and that that staff in hospitals making info available to the potential participants are not actively trying to recruit.
- Amend section on data storage. Anonymised data is to be stored in electronic encrypted form for 10 years.
- Consider also requesting assent from children (through their parents) this may require preparing a child-friendly information sheet that can be used by parents when explaining the study to their children.

Appendix 7: Study invitation letter

Hospital headed paper logo

Parent(s) of X

Re: Improving Outcomes for Children with Epilepsy (IOCE) Study.

Dear Parent(s),

I am writing to invite you to participate in the above research study, which we are carrying out in collaboration with University College Cork and Epilepsy Ireland. I have enclosed a study information sheet and a consent form. If you are happy to consent to be part of the research study, please return in the stamped addressed envelope enclosed.

We believe this research will enhance our knowledge of the needs of the children and families attending our service. Thank you for taking the time to read this letter and the enclosed.

Yours sincerely,

_____

Dr X
Consultant Paediatric Neurologist

Appendix 8: Study Information Sheet

Improving Outcomes for Children with Epilepsy (IOCE)

Study Information Sheet

What is the Study about?
Some children with epilepsy cope very well but some can struggle with the condition. This study will look at outcomes (medical, behavioural, school) in children who have epilepsy and at what medical, personal and family factors help or hinder the child in adjusting well to epilepsy. In addition, some participants will be invited to participate in a new intervention programme aimed at bolstering family resilience in order to improve outcomes for your child.

What will the Study involve?
Being part of the study will involve you completing some questionnaires about you and your child. The questionnaires will focus on topics such as how your child behaves and feels, how they are doing at school, what your family is like, your child's medical history, current seizure frequency and quality of life. The questionnaires, which you will access if you consent to participation in the study, take no longer than 40 minutes to complete.

We would also like to contact your child's school to see how they are getting on in school. This will involve asking your child's teacher to complete a similar questionnaire to the one you will complete if you consent to taking part in the research. Teachers will only be informed that a child in their class (your child) is participating in a research project at UCC. They will not be given information about your child's diagnosis.

Out of all the parents who consent to take part and complete the questionnaires, up to 20 will be invited to participate in a one-day intervention workshop in Cork City. The workshop will be delivered by Psychologists and is designed to help parents promote adjustment in their child and help them cope well with epilepsy. Similar interventions have proven beneficial to families of children with other chronic medical conditions and we wish to explore whether benefits may also be gained for families of children with epilepsy. Those who are eligible to take part in the one-day workshop will also be asked to complete questionnaires three months after the workshop.

Why have you been asked to take part?

You have been asked to be a part of this study as your child has a clinical diagnosis of epilepsy and is attending paediatric services in Cork.

Do you have to take part?

Participation is voluntary. If you agree to participate, we will ask you to sign a consent form. However, even if you have signed the consent form, you can still change your mind and withdraw from the study without need of explanation up to one month after participation. This study is entirely separate from your child's routine treatments and if you decide not to participate in the study, it will **not** affect your child's access to, or quality of, their treatment received.

Will your participation in the study be kept confidential?

Yes. We will ensure that no personally identifiable information is contained in any written reports about this study. Your name will be assigned a unique number and no personally identifying details will be used. We will keep the list of names and assigned numbers in a safe and secure password protected file. All information you give us will be treated confidentially unless you tell us something which suggests that your child or family are at risk of harm. This is the normal limit to confidentiality and only in such a situation would we have to talk with you about taking action to reduce risk.

What will happen to the information which you give?

The data will be kept confidential for the duration of the study, available only to the research team. All data will be stored in password protected files on encrypted computers. On completion of the project, data will be retained for a further 10 years and then destroyed. We guarantee that we will keep any information you provide confidential.

What will happen to the results?

The results will be presented in academic work which will be seen by the research team and academic assessors. This work may be read by future students. We also hope that the study will be published in a research journal and presented at conferences. You will also receive a summary of the study findings once complete.

What are the possible advantages and disadvantages of taking part?

Whilst those eligible for the intervention part of the study may very well gain some benefit, for the most part this study is about furthering our understanding of the psychological and family needs of children with epilepsy. Thus it may benefit future service provision rather than you directly. However, all participating families will receive a summary of the research findings which may be of interest and benefit.

We do not envisage any negative consequences for you in taking part in this study. However, it is possible that talking about your experiences may cause some distress. If you experience any unease as a result of being involved in the study, you will be able to contact Emma (emma.hennessy@hse.ie) or Sinead (sinead.hartley1@hse.ie) to discuss.

Who has reviewed this study?

The study has been approved by the School of Applied Psychology, UCC.

Any further queries?
If you would like to discuss any aspect of this study further before making a decision to participate, please feel free to contact Emma Hennessy (emma.hennessy@hse.ie) or Sinead Hartley (sinead.hartley1@hse.ie). Otherwise, please complete the enclosed consent forms and return them in the stamped addressed envelope provided.

The research team carrying out this work includes Emma Hennessy and Sinead Hartley (Doctoral students in Clinical Psychology, UCC); Dr Christopher McCusker (Consultant Paediatric Clinical Neuropsychologist and Senior Lecturer in Clinical Psychology); Dr Niamh McSweeney and Dr Olivia O' Mahony (Consultant Paediatric Neurologists, Cork University Hospital); and Niamh Jones and Loretta Kennedy (Epilepsy Ireland).

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Appendix 9: Study Consent Form

Improving Outcomes for Children with Epilepsy (IOCE) Study Consent Form

Please tick the boxes to indicate your consent

I, the undersigned, give my informed consent to:

1. Take part in the study ☐

2. Be contacted by Emma Hennessy and Sinead Hartley regarding the study ☐

3. For one of the named researchers to have access to my child's medical records so as to obtain details about their epilepsy ☐

4. For a researcher to contact my child's school to obtain information on my child's academic performance ☐

Once I have signed this consent I understand that:

- I am confirming that I have read and understood this form and the attached study information sheet.

- I will be assigned a unique number, under which all of my data will be stored confidentially.

- I am participating voluntarily and I may withdraw my participation without repercussions. I can do this by emailing Emma or Sinead within one month of submitting my questionnaires. If this happens I understand that my data from questionnaires will be deleted.

- I may be invited to take part in a one-day workshop.

- Any data obtained through my participation in this research will be treated as confidential and processed only in accordance with the Data Protection Acts, and that they will be used only for the purposes of research.

Please tick the box if you would like to receive the results of the study when it is completed ☐

Please sign here

Full Name: _____

Signed: _____ **Date:** _____

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Appendix 10: Demographic Questionnaire

Parent Information

1. Name of parent (1):

_____

2. Address:

_____

_____

_____

3. Telephone number: **4. Email address:**

_____ _____

5. Age: **6. Gender:** **7. Country of birth:**

_____ _____ _____

8. Marital status:

Single: ☐ Married: ☐ Separated: ☐ Divorced: ☐

9. Employed:

Yes: ☐ No: ☐

10. What is your highest level of qualification (please tick)?

Junior Certificate ☐ Bachelor's Degree ☐

Leaving Certificate ☐ Masters ☐

Certificate ☐ Doctorate ☐

Diploma ☐

11. Name of parent (2):

_____

12. Address:

_____

_____

_____

13. Telephone number: **14. Email address:**

_____ _____

15. Age: **16. Gender:** **17. Country of birth:**

_____ _____ _____

18. Marital status:

Single: ☐ Married: ☐ Separated: ☐ Divorced: ☐

19. Employed:

Yes: ☐ No: ☐

20. What is your highest level of qualification (please tick)?

Junior Certificate ☐ Bachelor's Degree ☐

Leaving Certificate ☐ Masters ☐

Certificate ☐ Doctorate ☐

Diploma ☐

21. Does either parent have a current illness or diagnosis?

Yes: ☐ No: ☐
If yes, please state:

_____
_____
_____

Family Composition

22. Are both parent's living in the same household?
Yes: ☐ No: ☐
23. Number of children in family:
_____
24. Ages of children in family:

- _____
- _____
- _____
- _____

25.

- _____
- _____
- _____
- _____

Do any of your other children have a current illness or diagnosis?

Yes: ☐ No: ☐
If yes, please state:

_____
_____
_____

26. Please tick which parent completed this questionnaire:
Parent 1: ☐ Parent 2: ☐

Appendix 11: Medical Questionnaire

Child Information

1. Name of child with Epilepsy:

_____

2. Age: **3. Gender:**

_____ _____

4. Does your child attend mainstream schooling (please tick)?

Yes: ☐ No: ☐

5. Name of school: **6. Name of Teacher:**

_____ _____

Epilepsy Information

7. Age at first seizure: **8. Age at diagnosis:**

_____ _____

9. Number of seizures in the past:

3 months: _____ 6 months: _____ 1 year: _____

10. Does your child have any other diagnoses/illnesses?

Yes: ☐ No: ☐

If yes, please state:

_____

Use of Anti-Epileptic Medication

11. Is your child currently taking any anti-epileptic medication (please tick)?

Yes: ☐ No: ☐

If yes, please state name of medication:

_____

Appendix 12: Battery of Psychometric Questionnaires

Section A

The following statements are about your child's behaviour. You are to decide which of those statements are NOT TRUE, SOMEWHAT TRUE or CERTAINLY TRUE for your child. Please then tick the appropriate column. Please answer based on your child's behaviour over the past six months or this school year. *There are no right or wrong answers.*

		NOT TRUE	SOMEWHAT TRUE	CERTAINLY TRUE
1.	Considerate of other people's feelings			
2.	Restless, overactive, cannot stay still for long			
3.	Often complains of headaches, stomach aches, or sickness			
4.	Shares readily with other children (treats, toys, pencils etc.)			
5.	Often has temper tantrums or hot tempers			
6.	Rather solitary, tends to play alone			
7.	Generally obedient, usually does what adults request			
8.	Many worries, often seems worried			
9.	Helpful if someone is hurt, upset or feeling ill			
10.	Constantly fidgeting or squirming			
11.	Has at least one good friend			
12.	Often fights with other children or bullies them			
13.	Often unhappy, down-hearted or tearful			
14.	Generally liked by other children			
15.	Easily distracted, concentration wanders			
16.	Nervous or clingy in new situations, easily loses confidence			
17.	Kind to other children			
18.	Often lies or cheats			
19.	Picked on, or bullied by other children			
20.	Often volunteers to help others (parents, teachers other children)			
21.	Thinks things out before acting			
22.	Steals from home, school or elsewhere			
23.	Gets on better with adults than with other children			
24.	Many fears, easily scared			
25.	Sees tasks through to the end, good attention span			

Section B

1. Please list the sports your child most likes to take part in. For example: swimming, baseball, skating, skate boarding, bike riding, fishing, etc.

☐ None

a. _____

b. _____

c. _____

Compared to others of the same age, about how much time does he/she spend in each?

	Less Than Average	Average	More Than Average	Don't Know
a.	☐	☐	☐	☐
b.	☐	☐	☐	☐
c.	☐	☐	☐	☐

Compared to others of the same age, how well does he/she do in each one?

	Below Average	Average	Above Average	Don't Know
a.	☐	☐	☐	☐
b.	☐	☐	☐	☐
c.	☐	☐	☐	☐

2. Please list your child's favourite hobbies, activities and games, other than sports. For example: stamps, dolls, books, piano, crafts, cars, computers, singing, etc. (Do ***not*** include listening to radio or TV).

☐ None

a. _____

b. _____

c. _____

Compared to others of the same age, about how much time does he/she spend in each?

	Less Than Average	Average	More Than Average	Don't Know
a.	☐	☐	☐	☐
b.	☐	☐	☐	☐
c.	☐	☐	☐	☐

Compared to others of the same age, how well does he/she do in each one?

	Below Average	Average	Above Average	Don't Know
a.	☐	☐	☐	☐
b.	☐	☐	☐	☐
c.	☐	☐	☐	☐

3. Please list any organisations, clubs, teams, or groups your child belongs to.

☐ None

a. _____

b. _____

c. _____

Compared to others of the same age, about how much time does he/she spend in each?

	Less Than Average	Average	More Than Average	Don't Know
a.	☐	☐	☐	☐
b.	☐	☐	☐	☐
c.	☐	☐	☐	☐

4. Please list any jobs or chores your child has. For example: paper route, babysitting, making bed, working in store, etc. (include both paid and unpaid jobs in chores.

☐ None

a. _____

b. _____

c. _____

Compared to others of the same age, about how much time does he/she spend in each?

	Less Than Average	Average	More Than Average	Don't Know
a.	☐	☐	☐	☐
b.	☐	☐	☐	☐
c.	☐	☐	☐	☐

5. About how many close friends does your child have? (Do *not* include brothers & sisters)

☐ **None** ☐ **1** ☐ **2 or 3** ☐ **4 or more**

6. About how many times a week does your child do things with any friends outside of regular school hours? (Do *not* include brothers & sisters)

☐ **Less than 1** ☐ **1 or 2** ☐ **3 or more**

7. Compared to others of his age, how well does your child:

	Worse	**Average**	**Better**	
a. Get along with his/her brothers & sisters?	☐	☐	☐	☐ Has no brothers or sisters
b. Get along with other kids?	☐	☐	☐	
c. Behave with his/her parents?	☐	☐	☐	
d. Play and work alone?	☐	☐	☐	

8. Performance in academic subjects.

Check a box for each subject that child takes	**Failing**	**Below Average**	**Average**	**Above Average**
a. Reading, English, or Language arts	☐	☐	☐	☐
b. History or Social Studies	☐	☐	☐	☐
c. Arithmetic or Math	☐	☐	☐	☐
d. Science	☐	☐	☐	☐
e. _____	☐	☐	☐	☐
f. _____	☐	☐	☐	☐
g. _____	☐	☐	☐	☐

Other academic subjects, for example: computer courses, foreign languages, business. Do *not* include gym, shop, driver's ed., or non-academic subjects.

9. Does your child receive special education or remedial services or attend a special class or special school? ☐ No ☐ Yes – kind of services, class, or school:

10. Has your child repeated any grades? ☐ No ☐ Yes – grades and reasons:

11. Has your child had any academic problems in school? ☐ No ☐ Yes – please describe:

_____

12. When did these start? _____

13. Have these problems ended? ☐ No ☐ Yes – when?

14. Does your child have any illness or disability (either physical or mental)?	☐ No ☐ Yes – please describe: _____
15. What concerns you most about your child?	
16. Please describe the best things about your child.	

Section C

The following questions relate to the impact that having epilepsy has on your child's quality of life. Please answer the questions thinking about the past **ONE MONTH**, and how much difficulty your child has had with each statement.

Impact		**Never**	**Almost Never**	**Sometimes**	**Often**	**Almost Always**
1.	My child has trouble doing the same physical activities or sports as other kids	0	1	2	3	4
2.	My child has trouble being as independent in daily tasks (e.g. dressing) as other kids his/her age	0	1	2	3	4
3.	My child's activities are restricted due to epilepsy	0	1	2	3	4
4.	My child has trouble taking his/her epilepsy medicine or doing other treatments (e.g. a special diet)	0	1	2	3	4
5.	My child has trouble avoiding seizure triggers (e.g. flashing lights, being tired)	0	1	2	3	4
6.	My child misses school or social activities because of epilepsy and/or its treatments	0	1	2	3	4
7.	My child does not like being left alone in case he/she has a seizure	0	1	2	3	4
8.	My child feels different from other kids or family members	0	1	2	3	4
9.	My child feels embarrassed when a seizure happens	0	1	2	3	4
Cognitive Functioning		**Never**	**Almost Never**	**Sometimes**	**Often**	**Almost Always**
1.	My child has trouble thinking quickly	0	1	2	3	4
2.	My child has trouble remembering things	0	1	2	3	4
3.	My child has trouble learning new things	0	1	2	3	4
4.	My child needs extra help at school	0	1	2	3	4
5.	My child has trouble understanding what he/she reads	0	1	2	3	4
6.	My child has trouble keeping up with school work	0	1	2	3	4
Sleep/Fatigue		**Never**	**Almost Never**	**Sometimes**	**Often**	**Almost Always**
1.	My child feels tired during the day	0	1	2	3	4
2.	My child has trouble sleeping (e.g. falling and staying asleep)	0	1	2	3	4
3.	My child needs more sleep than other kids	0	1	2	3	4
Executive Functioning		**Never**	**Almost Never**	**Sometimes**	**Often**	**Almost Always**
1.	My child has trouble sitting still	0	1	2	3	4
2.	It is hard for my child to do what he/she is told	0	1	2	3	4
3.	My child has trouble paying attention	0	1	2	3	4
4.	My child has trouble finishing things he/she has started	0	1	2	3	4
5.	My child acts without thinking	0	1	2	3	4

6.	My child has trouble staying organised	0	1	2	3	4
Mood/Behaviour		**Never**	**Almost Never**	**Sometimes**	**Often**	**Almost Always**
1.	My child feels grouchy	0	1	2	3	4
2.	My child feels angry	0	1	2	3	4
3.	My child feels sad or blue	0	1	2	3	4
4.	My child feels *afraid or scared/ tense or anxious	0	1	2	3	4
5.	My child is easily frustrated	0	1	2	3	4

*** If your child is aged between 8-12yrs, please use afraid or scared when answering. If your child is aged between 13-18 yrs., please use tense or anxious when answering.**

Section D

The following statements are about families. You are to decide which of those statements are TRUE or MOSTLY TRUE for your family and which are FALSE or MOSTLY FALSE. Please then tick the appropriate column. *There are no right or wrong answers.* You may feel that some of the statements are true for some family members and false for others. Decide which is the stronger OVERALL impression and answer accordingly. We are interested in YOUR impression. Do not try to figure out how other members of your family see it, but do give us your general impression for each statement.

		TRUE	**FALSE**
1.	Family members really help and support one another.		
2.	Family members often keep their feelings to themselves.		
3.	We fight a lot in our family.		
4.	We often seem to be killing time at home.		
5.	We say anything we want to around home.		
6.	Family members seldom become openly angry.		
7.	We put a lot of energy into what we do at home.		
8.	It's hard to blow off steam at home without upsetting somebody.		
9.	Family members sometimes get so angry they throw things.		
10	There is a feeling of togetherness in our family.		
11	We tell each other about our personal problems.		
12	Family members hardly ever lose their tempers.		
13	We seldom volunteer when something has to be done at home.		
14	If we feel like doing something on the spur of the moment we just pick up and go.		
15	Family members often criticise each other.		
16	Family members really back each other up.		
17	Someone usually gets upset if you complain in our family.		
18	Family members sometimes hit each other.		
19	There is little group spirit in our family.		
20	Money and paying bills is openly talked about in our family.		
21	If there's a disagreement we try to smooth things over and make the peace.		
22	We really get along well with each other.		
23	We are usually careful about what we say to one another.		
24	Family members often try to one-up or out-do each other.		
25	There is plenty of time and attention for everyone in our family.		
26	There is a lot of spontaneous discussion in our family.		
27	In our family we believe you don't ever get anywhere by raising your voice.		

Section E

Please read each statement carefully. Decide how well you believe it describes your family now from your view point. Please then tick the appropriate column. Your "family" may include any individuals you wish.

		Strongly Agree	Agree	Disagree	Strongly Disagree
1	Our family structure is flexible to deal with the unexpected				
2	Our friends value us and who we are				
3	The things we do for each other make us feel a part of the family				
4	We accept stressful events as part of life				
5	We accept that problems occur unexpectedly				
6	We all have input into major family decisions				
7	We are able to work through pain and come to an understanding				
8	We are adaptable to demands placed on us as a family				
9	We are open to new ways of doing things in our family				
10	We are understood by other family members				
11	We ask neighbours for help and assistance				
12	We attend church/synagogue/mosque services				
		Strongly Agree	**Agree**	**Disagree**	**Strongly Disagree**
12	We believe we can handle our problems				
14	We can ask for clarification if we do not understand each other				
15	We can be honest and direct with each other in our family				
16	We can blow off steam at home without upsetting someone				
17	We can compromise when problems come up				
18	We can deal with family differences in accepting a loss				
19	We can depend upon people in this community				
20	We can question the meaning behind questions in our family				
21	We can solve major problems				
22	We can survive if another problem comes up				
23	We can talk about the way we communicate in our family				
24	We can work through difficulties as a family				
25	We consult with each other about decisions				
26	We define problems positively to solve them				
27	We discuss problems and feel good about the solutions				
		Strongly Agree	**Agree**	**Disagree**	**Strongly Disagree**
28	We discuss things until we research a resolution				
29	We feel free to express our opinions				

34	We feel we are strong in facing big problems				
35	We have faith in a supreme being				
36	We have the strength to solve our problems				
37	We keep our feelings to ourselves				
38	We know there is community help if there is trouble				
39	We know we are important to our friends				
40	We learn from each other's mistakes				
41	We mean what we say to each other in our family				
42	We participate in church activities				
		Strongly Agree	**Agree**	**Disagree**	**Strongly Disagree**
43	We receive gifts and favours from neighbours				
44	We seek advice from religious advisors				
45	We seldom listen to family concerns or problems				
46	We share responsibility in the family				
47	We show love and affection for family members				
48	We tell each other how much we care for one				
49	We think this is a good community to raise children				
50	We think we should not get too involved with people in this community				
51	We trust things will work out even in difficult times				
52	We try new ways of working with people				
53	We understand communication from other family members				
54	We work to make sure family members are not emotionally or physically hurt				
55	Is there something else which helped your family through this adverse event that has not been described or discussed				

Section F

The following statements are about worries that some parents of children with chronic illnesses have. Please indicate how much you worry about each of the following by circling the appropriate number alongside each statement. Again there are no right or wrong answers.

		1 Not at all	**2 Sometimes**	**3 Often**	**4 Most of the time**
1.	I worry that my child will look different from other teenagers or adults because of his/her health condition.	1	2	3	4
2.	I worry that my child will have a harder time finding a boyfriend or girlfriend because of the health condition.	1	2	3	4
3.	I worry that my child won't get married because of his/her health condition.	1	2	3	4
4.	I worry that my child will get worse or will get very sick again.	1	2	3	4
5.	I worry that my child won't be able to do things he or she wants to do because of the health condition.	1	2	3	4
6.	I worry that my child will have a hard time getting around or going places compared to other teens or adults.	1	2	3	4
7.	I worry that my child will need medications or will need stronger medications.	1	2	3	4
8.	I worry that my child will always have to take medication.	1	2	3	4
9.	I worry that my child will have future side-effects from his/her medications.	1	2	3	4
10.	I worry that my child will grow up too fast because of the health condition.	1	2	3	4
11.	I worry that my child won't be able to handle things in the future when she/he is on his/her own.	1	2	3	4

Appendix 13: Extended Methodology

The current study utilised a cross-sectional survey style research design. By employing a survey style design, the authors hoped that they would be able to access as large a sample as possible within the designated region, and within the resources that were available to them, so as to create as accurate a profile as possible of risk and protective factors in relation to psychological family factors and psychosocial outcomes for children with epilepsy in the Munster region.

Survey Design

The current survey was made up of a battery of standardised psychometric questionnaires, as well as two bespoke questionnaires which were developed to gather demographic and medical/illness related information. The standardised questionnaires were chosen by the authors based on the rationale, and research questions, outlined in section 1 of this paper.

The survey was paper, and pencil based and was sent to participants via standard post. The survey pack also included a stamped addressed envelope for ease of return. In total, it took approximately 40 minutes to complete from start to finish.

Survey Strengths

There are a number of strengths and advantages to utilising surveys in research. For example, surveys are relatively inexpensive and so are capable of accessing large samples in a relatively short period of time, which in turn can allow for more generalisable findings. Surveys are typically non-invasive and allow people to express their true attitudes/opinions perhaps more easily than they could in a face-to-face setting, where participants may wish to portray themselves in a 'socially desirable' manner so as not to violate any perceived social norms. Still, it must be noted that surveys cannot completely account for such social desirability effects and the researcher must remain cognisant that such effects could indeed be present in their research. However, completing the current survey in the privacy of their own homes likely made it easier for participants to be more honest in their responses than if the survey was administered to them by the researcher in person.

Further, surveys can be completed by participants from the comfort of their own home and so can be completed within their own time. By facilitating home completion and providing a method of return, surveys potentially allow researchers to access participants who may not be able to come to meet individually due to already busy lives and who have multiple time constraints.

Survey Limitations

Although there are certainly many identifiable strengths to the use of survey style designs within research, there are also a number of limitations which are worthy of consideration.

Firstly, the issue of causal relations is likely the most important factor to be aware of when considering survey style research. The question of causality cannot be tested

definitively in cross-sectional designs and one cannot conclusively interpret relationships between variables in a particular direction. Rather, relationships between variables are more likely to be bi-directional in nature and so longitudinal research is needed to address this issue and draw more accurate interpretations over time.

A second limitation is that of informant bias. Much research, including the current study, utilises a single informant to report on multiple perspectives, for example a mother reporting on her own experiences, her family's experiences and her child's experiences. Although in the current study the majority of questionnaires were completed by the child with epilepsy's mother, the study also collected data from the child's teacher, who represented a non-biased informant for comparison.

Lastly, surveys assume a certain level of literacy from participants. This may limit people with lower levels of literacy for effectively participating in research where their voice and viewpoint is valued, particularly if the survey is very lengthy. It may also lead to item nonresponse where participants chose to only answer certain questions on the survey. Providing an option for participants to receive support when completing the survey can help eliminate this risk. However, this was not possible due to resource constraints in the current study.

Appendix 14: Supplementary Table 2

Supplementary Table 2: Comparison of mean scores for parent and teacher-reported outcome scores for the SDQ, including effect size deviations

Scale	Parent report (n = 48) Mean ± SD	% in clinical range	Teacher report (n = 27) Mean ± SD	% in clinical range	Cohen's d
Prosocial	7.6 ± 2.43	25	8.1 ± 2.11	11	.2*
Peer difficulties	2.5 ± 1.92	48	.9 ± 1.37	7	-.9***
Hyperactivity/inattention	4.1 ± 2.52	23	3.6 ± 2.19	18	-.2*
Conduct	2.5 ± 2.21	44	.5 ± 1.12	7	-1.0***
Emotional difficulties	4.7 ± 2.82	65	3.2 ± 2.59	26	-.5**
Total difficulties	13.9 ± 6.67	50	8.3 ± 5.41	30	-.9***

SDQ = Strengths and Difficulties Questionnaire, SD = standard deviation
*small effect size, **medium effect size, ***large effect size