The Psychosocial Impact of Epilepsy on Children and their Families: How Clinicians can Provide Support

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> "Physicians may learn more from their patients than vice versa, and our experiences with the families of children with Dravet syndrome illustrate this phenomenon."
>
> (Nolan et al., 2008)

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Chapter 1

Psychosocial Interventions for Children and Young People with Epilepsy: A Repeat Systematic Review of the Literature in 2023

Prepared in accordance with the author requirements for 'Epilepsy and Behaviour'

Link (Full 2023 Author Guidelines): https://osf.io/bwnr2

Abstract

Background

Epilepsy is a lifelong neurological disorder that has a profound impact on the lives of millions of children and young people throughout the world, and is linked with mental ill-health and a poorer quality of life. Psychosocial interventions have showed promise for children and young people with epilepsy (CYPE), however there is an absence of large-scale RCT's that would add robustness to the evidence base. The present systematic review provides an update and extension of findings from an earlier review conducted in 2014 by Corrigan et al. (2016) to assess the state of the literature in 2023.

Methods

The present systematic review carried out a search of six electronic databases. Forward and backward chaining was carried out on review articles as well as the studies returned through the search to source additional studies. In total, ten articles were included in this review and appraised for quality using the Crowe Critical Appraisal Tool (CCAT) (Crowe, 2013).

Results

Forty percent (4/10) of the included studies were rated as high quality according to the CCAT, which represents a significant proportional increase since Corrigan et al.'s (2016) review. A meta-analysis of results was not possible due to significant methodological heterogeneity, and the variability of outcome measures, however effect sizes were reported or calculated for the majority of studies (7/10), which facilitated comparison. Despite the issues of relatively small samples, there are promising findings with regard to psychosocial interventions increasing epilepsy knowledge, coping strategies, self-efficacy, and quality of life markers.

Conclusions

There is a growing evidence base supporting the efficacy of psychosocial interventions for children and young people with epilepsy. This evidence base is also increasing in quality. Particular components of treatment that prove to be effective include psychoeducation, components based on cognitive behavioural therapy principles, as well as mindfulness techniques. This aligns with the evidence-based recommendations for adult populations. Intervention goals centre around improving quality of life, reducing symptom distress, and increasing knowledge and skills. The instruments used to measure these outcomes are predominantly standardised, however remain heterogeneous between studies which impacts the overall robustness of the evidence base.

1. Introduction

The present systematic review provides an update and extension of findings from an earlier review conducted in 2014 by Corrigan et al. (2016).

1.1 Background and Prevalence

Epilepsy is a lifelong neurological disorder that has a profound impact on the lives of millions of children and adolescents throughout the world. It has a reported prevalence of between 0.5 and 1%, and is the most present chronic neurological condition in children and adolescents (Aaberg et al., 2017).

1.2 Seizure Prevention

Seizure prevention is an important aspect of care for many people with epilepsy, and this is managed predominantly through pharmaceutical interventions. These interventions are considered successful for the majority of patients from either the first or second anti-epileptic medication trialled (Brodie et al., 2012). For those who do not respond favourably to the first two trials of medication, there are alternatives that have a proven evidence base, such as the ketogenic diet (see Martin-McGill et al., 2020 for a systematic review), and for those with disabling focal onset seizures surgical resection can be considered. 'Seizure freedom' has been found to be highly correlated with an improved quality of life in people with epilepsy (Birbeck et al., 2002). Despite successful methods of treatment, in a recent journal article, the authors state that 'epilepsy is one of the most common and disabling neurologic conditions, yet we have an incomplete understanding of the detailed pathophysiology and, thus, treatment rationale for much of epilepsy.' (Nazarov, 2022).

1.3 Psychosocial Issues Associated with Epilepsy

Beyond the psychopharmaceutical needs of people with epilepsy, there are often also associated psychological and psychosocial challenges. These can have several causes from an acute fear response associated with seizures (Biraben et al., 2001), to the avoidance of activities associated with positive mental health such as exercise, due to epilepsy-related fears (see Johnson et al., 2020 for a review).

A very large (36,984 participant) population-based study (Tellez-Zenteno et al., 2007) found that the lifetime prevalence for any mental health disorder for those without epilepsy was 20.7% (95% CI = 19.5–20.7), whereas people with epilepsy faced a significantly increased risk of mental health disorders across their lifespan (35.5%, 95% CI = 25.9–44.0). Depressive disorders have been found to be the mental health disorder with the highest co-morbidity with epilepsy (Kanner & Balabanov, 2002). People with a diagnosis of epilepsy are more likely to experience suicidal ideation (25%) across their lifespan, compared to those without epilepsy (13.3%) (Tellez-Zenteno et al., 2007). In a recent systematic review of the literature, Lu et al. (2021) found a high prevalence for mood disorders (35%) and anxiety disorders (25.6%) among adults with epilepsy.

In their large sample (n=250) study, Pham et al. (2017) found that people with epilepsy were significantly more likely to experience anxiety, and that this was associated with several negative outcomes, including a lower quality of life. Even when isolating other factors, such as frequency and severity of seizures, mental health remains a significant predictor of quality of life in adolescents with epilepsy (Healy et al., 2020), highlighting a need for evidenced psychological interventions. In their qualitative study, Fayed et al. (2021) found that the main theme with regard to dealing with the anxiety and uncertainty caused by epilepsy for adolescents was "to adapt or not to adapt" with the subthemes of this being "leave me alone" versus "sharing knowledge, empowering self". This emphasises the role for psychosocial interventions for children and young people with epilepsy (CYPE) to engage them and support the development of adaptive strategies to meet the challenges that they face.

The goals associated with any intervention for CYPE should address these factors, namely attitudes towards seizures, issues with mental health, deterioration in quality of life, and training / skill learning needs.

1.4 Psychosocial Interventions for People with Epilepsy

Psychosocial interventions have consistently been shown to be effective in reducing depression symptoms within the child and adolescent population, albeit with questions arising relating to the longevity of the treatment effect (see Watanabe et al., 2007 for a review). This is also the case for anxiety (see James et al's., 2020 Cochrane review). As

CYPE experience elevated levels of depression and anxiety, this suggests a role for psychosocial interventions within this population. This need is supported by the evidence-based recommendations for psychological treatments for people with epilepsy (Michaelis et al., 2018b).

The NICE guidelines (2022) recommend ongoing clinical discussions about the cognitive and mental health challenges that children and young people face, which can be associated with their epilepsy and/or treatment. In addition to this, there is specific mention within the NICE guidelines of the common neurobehavioural disorders that are frequently comorbid with epilepsy, such as attention deficit hyperactivity disorder (ADHD) and autism spectrum disorder (ASD). The prevalence of ADHD and ASD amongst people with epilepsy adds to the case for psychological interventions, and may require specific modifications to the delivery of these interventions. A diagnosis of ASD, for example, is associated with a higher risk of depression (see DeFilippis, 2018 for a review for adolescent population), however therapeutic work must be cognisant of the challenges presented with a poorer self-recognition of emotional states within the ASD population (Lainhart & Folstein, 1994) as well as challenges with cognitive flexibility (Mazefsky & White, 2014). In their Cochrane review of psychotherapy (predominantly CBT) for anxiety, however, James et al. (2020) found no significant differences in treatment effects between ASD and non-ASD populations.

Previous research has suggested that epilepsy is more prevalent in areas of social deprivation within England (Steer et al., 2014), as well as among people with lower incomes within the United Kingdom (Ferro, 2011). Such findings present context for psychological treatment work with people with epilepsy. Indeed, a recent prospective population-level study in Scotland concluded that "There is a clear social gradient to the incidence of early childhood epilepsies", suggesting significantly higher incidence of childhood epilepsy in areas of social deprivation (Symonds et al., 2021).

1.5 Research Landscape

The earlier review by Corrigan et al. (2016) noted that psychological interventions for people with epilepsy was a growing area of research, however that the primary focus was on adult populations. Michaelis & colleagues have recently provided several important

Cochrane systematic reviews in this area (Michaelis et al., 2018b; Michaelis et al., 2020) supporting the creation of evidence-based practice guidelines (Michaelis et al., 2018a). Whilst these reviews did include studies with child and adolescent populations, the vast majority of studies contained adult only populations (e.g., 75% within Michaelis et al., 2018b).

Systematic reviews have also evaluated the efficacy of specific treatment deliveries for the adult population, for example group self-management interventions (Smith et al., 2017), however this has not been extended to children and adolescents. Indeed, a more recent review concerning the child and adolescent population focussed on parenting interventions and parental outcomes (Kaye, 2021). Further research into the specific components of direct treatment that are most efficacious for CYPE would facilitate the development of evidence-based practice, improving outcomes for this population specifically. Research into the methods of delivery that yield the best results is also important, as remote delivery (e.g. teletherapy and online interventions) and group-based interventions offer cost savings when compared to individual psychotherapy, if found to be suitably efficacious.

We therefore believe that the present review represents a timely update to the previous review (Corrigan et al., 2016). We have set a lower methodological threshold for study inclusion than the Cochrane reviews (Michaelis et al., 2018b; Michaelis et al., 2020), allowing the inclusion of studies beyond randomised controlled trials (RCT's). The focus of this review is exclusively on the child and adolescent population, whereas this was a minority aspect of the Cochrane reviews. We will also focus on direct interventions that contain outcome data for the children and adolescents, rather than the indirect intervention focus used by Kaye (2021) or the narrower scope of care delivery and self-management strategies found in Fleeman et al.'s (2022) Cochrane review.

1.6 Definition of Psychosocial / Psychological Therapies

We have defined a psychosocial intervention in the following way for the purpose of this review: "A direct intervention that is primarily therapeutic, without a pharmacological or dietary-based element, that yields psychosocial outcome data (e.g. quality of life, reduction of distress)."

Studies can therefore include recognised evidence-based psychological interventions (e.g. Cognitive Behavioural Therapy, Acceptance and Commitment Therapy), as well as psychosocial interventions that develop communication skills, and health-education programmes that have direct delivery (i.e. to the child and adolescent population), and have psychosocial outcome measurements.

This would not include studies where the primary focus is cognitive rehabilitation, for example using computer software for attention retraining. Studies where the intervention was indirect (e.g. delivered to parents with parental outcomes) were also not included in the current review. Studies that did not yield psychosocial outcome data (e.g. psychoeducational interventions that only measure the assimilation of epilepsy-based knowledge) were not included.

1.7 Review Questions / Aims

The previous review (Corrigan et al., 2016) concluded that there was "limited but promising evidence that psychosocial interventions can be of benefit to CYPE improving mood, quality of life, and epilepsy knowledge. However, there is a need for further good quality studies using randomized controlled trial designs with larger samples." (Corrigan et al., 2016). The present review aims to evaluate the research evidence published since 2014, providing an overview of recent progress in this important area of healthcare for young people with chronic health problems.

The research questions and aims of the previous review (Corrigan et al., 2016) are retained to facilitate comparison between time periods. Minor alterations have been made to the wording of the questions.

1. Is there any evidence that psychosocial interventions are effective for children or young people with epilepsy?
2. Are there specific treatment components or methods of delivery that may increase the effectiveness of these interventions?
3. Are there clear intervention goals and how effectively are these measured?

2. Method

The present systematic review followed the PRISMA statement for guidance and structure throughout the process (Page et al., 2021) (Appendix A).

This review provides an update and extension to the systematic search conducted in 2014 by Corrigan et al. (2016). One key change is that the search strategy has been updated for greater sensitivity. The search strategy was developed to facilitate the inclusion of specific therapies that have been used for people with epilepsy within the search terms. The included therapeutic terms were taken from review articles (Lecce et al., 2023; Michaelis et al., 2018b; Michaelis et al., 2020).

2.1 Search Strategy

The electronic databases from the previous review (Corrigan et al., 2016) were retained in this review. This meant that the following were utilised Embase, Medline, and PsychInfo (via OVID online); CINAHL, and Psychology & Behavioral Sciences Collection (via EBSCO host); and Web of Science Core Collection (via Web of Knowledge). Final searches were conducted on 30th May 2023.

The search terms for the present review, as used for OVID: Medline and Embase is presented below.

1. exp Epilepsy
2. Epilep*.ti,ab,kw
3. 1 OR 2
4. exp child/
5. exp adolescent/
6. (Child* OR Adolescen* OR Young Person OR Young People OR Kids OR Minor* OR Youth* OR Paediatri* OR Pediatri*).ti,ab,kw
7. 4 OR 5 OR 6
8. ((Psychosocial OR Psychoeducation* OR Psycholog* OR Psychotherap* OR Mental Health) adj3 (Interven* OR Treat* OR Therap*)).ti,ab,kw
9. exp psychotherapy
10. exp cognitive therapy
11. exp cognitive behavioral therapy
12. (education program* OR behavioural strateg* OR behavioral strateg* OR motivational interviewing OR epilepsy education OR self-management OR cognitive behavioural therap* OR cognitive behavioral therap* OR CBT OR acceptance and commitment therapy OR ACT OR behavioural activation OR behavioral activation OR cognitive therap* OR cognitive restructuring OR stress management OR communication skills OR mindfulness).ti,ab,kw
13. 8 OR 9 OR 10 OR 11 OR 12
14. 1 AND 7 AND 13

A date range was also applied to the searches, so that only results published since the previous review were retuned. The date range for the present review succeeds the date range from the previous review (Corrigan et al., 2016), which were 1st January 1989 and 28th November 2014.

Limits were applied to the searches so that only English language studies (any territory) with human participants were returned.

Age filters were not applied to the search results, as the factor of age was addressed through the search strategy. The age ranges from the previous review (Corrigan et al., 2016) were retained, meaning that the World Health Organisation definition of 'adolescence' was adopted (World Health Organisation, 2023).

Duplicate studies were removed using the in-built tool within Microsoft EndNote, as well as through methodical manualised sorting.

Inclusion criteria:

- Published in English language
- Published in a peer-reviewed journal
- Studies published between 29th November 2014 and 30th May 2023
- Studies containing original data
- Intervention participants must be between ages of 0 and 19 years
- Intervention participants have a diagnosis of epilepsy

Exclusion criteria:

- Intervention participants are non-human
- Intervention participants do not have a diagnosis of epilepsy
- Intervention participants have a diagnosis of a learning disability
- Intervention participants are adult
- Studies without psychosocial intervention
- Studies without psychosocial outcome measurements

The following types of research article were also not included; qualitative studies, review studies (systematic, literature) and meta-analyses, case studies, conference abstracts or

presentations, book sections, and commentaries or opinion pieces. These were retained from the earlier review.

In addition to the systematic search, manual searches were carried out using the reference lists from selected reviews (Lecce et al., 2023; Michaelis et al., 2018b; Michaelis et al., 2020), as well as forward and backward chaining from the final included studies. This led to the inclusion of one additional study for full text review.

2.2 Quality Assessment

To facilitate comparison between the present review and the previous review (Corrigan et al., 2016), we retained the use of the Crowe Critical Appraisal Tool (CCAT) (Crowe, 2013), which remains in version 1.4. This quality assessment tool for systematic reviews supports the evaluation of included studies based on their reporting and methodology.

The previous review (Corrigan et al., 2016) transposed the scores obtained from applying the CCAT to the studies, which are out of 40, to percentages. Studies were then categorised according to their percentage score as shown in Table 1.1.

Table 1.1

CCAT Scoring Key

Quality Rating	Percentage	Equivalent CCAT Score
Poor Quality	≤50	20/40 or less
Acceptable Quality	51 - 74	21/40 to 29/40
High Quality	≥75	30/40 and above

We have retained this scoring and rating system for the present review so as to facilitate comparisons between the research landscapes at each time period. The transposition of scores to percentages is supported by the author of the CCAT (Crowe, 2013).

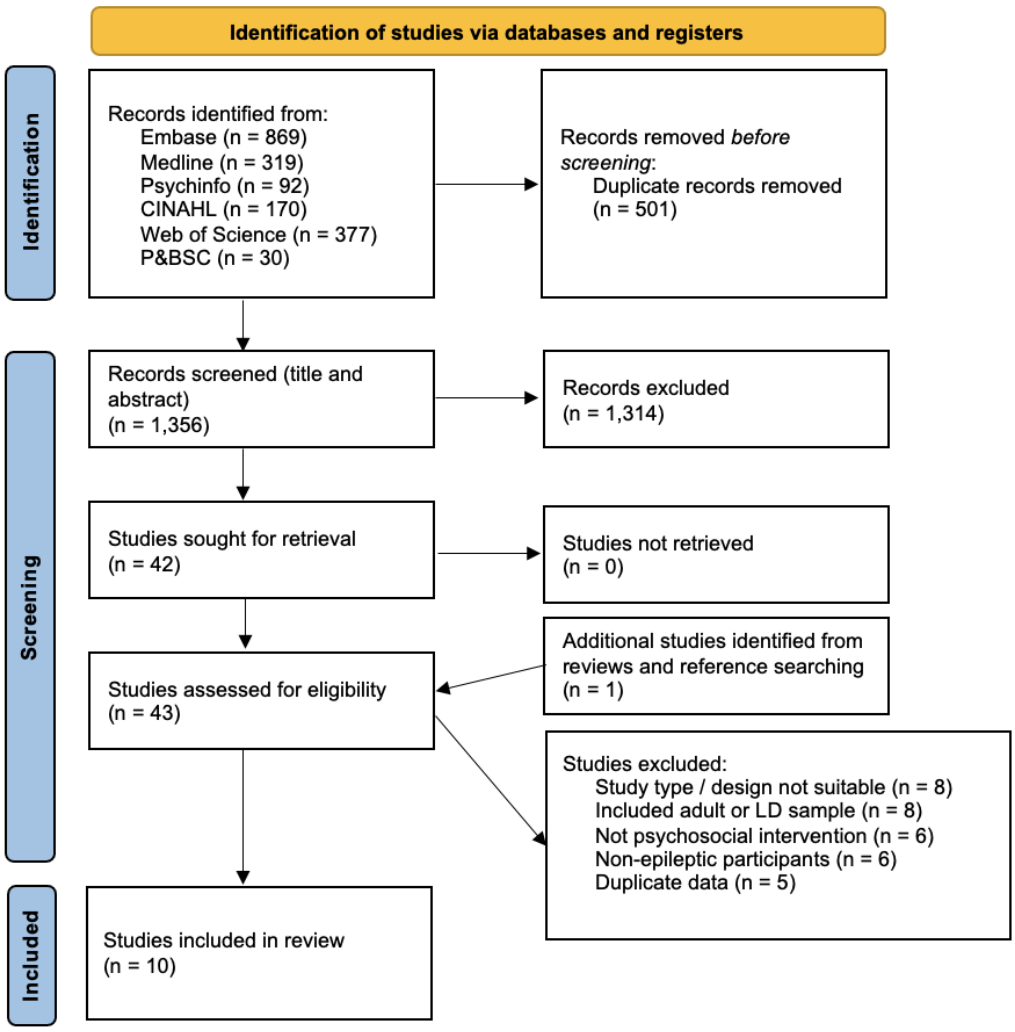
3. Results

3.1 Search Results

A random sample of 10% (136/1,356) of the records that were title and abstract screened for full text review were also blind rated by a second rater to determine inter-rater reliability. This returned an initial score of κ = 0.699 representing 'substantial agreement' (Landis & Koch, 1977). In practice, this related to four incidences of disagreement which were resolved through discussion.

Figure 1.1

PRISMA Diagram: Adapted from Page et al. (2021).



3.2 Quality Rating Results

Studies were assessed for quality using the CCAT. Table 1.2 details the scores for each study across the 8 domains of the CCAT, as well as the total score, a transposed percentage, and the associated quality rating.

All ten included studies were rated using the CCAT by the lead author and an additional blind-rater, thus forming a 'rating pair'. The rating pair had an initial agreement of $\kappa = 0.878$, which represented one disagreement (1/10). Weighted Cohen's kappa was used as the categories (CCAT classifications) were ordered. Using Landis & Koch's (1977) benchmarks, this represents 'almost perfect agreement'. After discussion between raters, perfect agreement was attained for all articles.

A summary of the ten studies included in the present review is provided in Table 1.3.

Table 1.2

CCAT Quality Ratings

Study	Preliminary	Introduction	Design	Sampling	Data Collection	Ethical Matters	Results	Discussion	Total Score (%)	Rating
Batista et al. (2015)	3	2	2	1	2	1	3	3	42.5	Poor
Brown et al. (2019)	5	5	3	3	3	4	4	5	80	High
Dorris et al. (2017)	4	5	4	4	4	4	4	5	85	High
Eom et al. (2016)	2	2	2	1	1	2	3	4	42.5	Poor
Gurhopur et al. (2018)	3	3	4	3	3	2	3	4	62.5	Acceptable
Guven et al. (2020)	4	5	4	5	4	3	4	4	82.5	High
Rizou et al. (2017)	3	3	3	3	2	3	2	4	57.5	Acceptable
Schaffer et al. (2017)	3	4	4	3	3	3	5	5	75	High
Svanstrom et al. (2023)	5	5	2	2	2	3	4	5	70	Acceptable
Tajrishi et al. (2015)	2	2	4	4	2	2	3	4	57.5	Acceptable

Table 1.3

Data Extraction: Summary of 10 Included Studies that Met Criteria

Study	Sample	Design	Intervention Delivery	Psychosocial Outcome Measures	Analysis	Main Findings (Child / Young Person)
Batista et al. (2015)	17 children, 9-17 years. Epilepsy diagnosis >1 year. Purposive sample. Croatia.	One group, pre-test/post-test. No follow-up.	Manualised computer-assisted CBT delivered in a residential setting. Team included paediatricians, psychologists, and nurses. CBT intervention had six modules; three on epilepsy education, three on coping strategies.	Scale of Coping with Stress (SUO) and author created two knowledge tests; one for general epilepsy knowledge, and one for epilepsy and coping.	Related samples Wilcoxon signed rank test.	Significantly higher epilepsy knowledge post-intervention ($p<0.01$). Significantly higher scores on stress knowledge and coping with stress quiz post-intervention ($p<0.01$). Significantly higher (min = $p<0.05$) usage frequency and effectiveness of 4 strategies on the SUO (problem solving, seeking help from friends, seeking help from family, and cognitive restructuring).
Brown et al. (2019)	115 children, 8-14 years. >1 seizure in past 12 months. Convenience sample. Canada.	RCT (conformed to CONSORT 2017 guidelines). 6-month follow-up period.	Intervention group had physical activity behaviour-change counselling, which were motivational and psychoeducational (self-regulatory skills).	Physical activity markers. Childhood epilepsy quality of life scale (CEQOL). KIDSCREEN-27 which measures health-related quality of life. The Children's depression inventory-short (CDI-S).	Linear regression model, independent t-tests, Chi-square.	No significant differences between groups for condition specific quality of life ($p >0.07$), health-related quality of life ($p>0.15$), or depressive symptoms ($p>.07$). No significant difference between groups for physical activity ($p = 0.67$).
Dorris et al. (2017)	83 children, 12-17 years. Epilepsy diagnosis >6 months. Convenience	RCT using a waiting list control group. Follow-up at three and six months.	Intervention delivered in groups by healthcare professionals (epilepsy nurse and clinical psychologist). Weekly sessions, manualised	Paediatric Quality of Life Inventory (PedsQL), Glasgow Epilepsy Outcome Scale for Young Persons (GEOS-YP), Epilepsy Knowledge Profile-General (EKP-G), the Seizure Self Efficacy Scale for Children (SSEC-C), the Brief -	T-tests, Mann-Whitney test, McNemar's test.	Significant increase in epilepsy knowledge in experimental group after intervention ($p = 0.04$, $d = 0.25$), and at three-month follow-up ($p = 0.02$, $d = 0.58$). Positive changes noted in GEOS-YP, BPIQ, PI-ED, and SSEC for intervention

	sample. United Kingdom.		delivery based on CBT and mindfulness techniques	Illness Representations Questionnaire (B-IPQ), Paediatric Index of Emotional Distress (PI-ED) as well as participant and caregiver questionnaires created for the study.		group, however these changes did not reach significance post-intervention or at follow-up.
Eom et al. (2016)	10 children, 8-12 years. Benign epilepsy diagnosis. Convenience sample. Korea.	One group, pre-test/post-test. No follow-up.	35-week exercise program. Gym and home-based. Parents received psycho-educational input from healthcare professionals, including clinical psychologists.	Korea–Child Behavior Checklist (K-CBCL) and the Korean version of the Quality of Life in Childhood Epilepsy Questionnaire (K-QOLCE).	Wilcoxon signed-rank test. Outliers were removed for some analyses.	Significant improvement in general health ($p = 0.018$, $g = 2.62$), and quality of life ($p = 0.017$, $g = 2.47$) post intervention on the QOLCE. A reduction in behaviour problems post-intervention, however non-significant (K-CBCL). No significant change was noted for competence post-intervention (K-CBCL).
Gurhopur et al. (2018)	92 children, 7-18 years. Epilepsy diagnosis >6 months. Convenience sample. Turkey.	RCT. Follow-up at one and three months.	Modular education program. Activities included discussions, brainstorming, Q&A, role playing, and playing games.	The Epilepsy Knowledge Test for Children (EKTC), the Seizure Self-efficacy Scale for Children (SSES-C), the Quality of Life in Epilepsy Inventory (QOLIE-48).	Chi-square, t-tests, Kolmogorov-Smirnov test.	Scores on the EKTC ($p<0.001$, $d = 0.92$), SSES-C ($p<0.001$, $d = 0.27$), and QOLIE-48 ($p<0.001$, $d = 0.34$) increased significantly post-intervention for the intervention group.
Guven et al. (2020)	69 children, 9-18 years. Epilepsy diagnosis >6 months. Convenience sample. Turkey.	RCT. No follow-up.	Access to a web-based epilepsy education program (WEEP) for 12 weeks. Sent weekly reminders to use the website.	Epilepsy Knowledge Test (EKT), Seizure Self-Efficacy Scale for Children (SSES-C), Child Attitude Toward Illness Scale (CATIS), the e-Health Literacy Scale (eHEALS).	Chi-square, t-tests.	Intervention group had statistically different post-test scores (within group) for all measures ($p<0.05$). EKT ($p = <0.0001$, $d = 1.32$). SSES-C ($p = <0.0001$, $d = 1.48$). CATIS ($p = <0.0001$, $d = 0.97$). eHEALS ($p = <0.0001$, $d = 1.01$).
Rizou et al. (2017)	24 children, 12-17 years. >1 seizure in past 12 months. Purposive sample. Greece.	Matched pairs design. Epilepsy control group.	Brief self-regulation-based intervention, one 4-hour group session. Psycho-educational component, relaxation, and storytelling.	Brief Illness Perceptions Questionnaire (BIPQ), Revised Children's Anxiety and Depression Scale (RCADS), Athens Insomnia Scale (AIS), and the somatization scale of the	ANCOVA	Significant main effects noted for psychological distress levels ($p = 0.005$, $g = 1.37$), and sleep problems ($p = 0.003$, $g = 1.83$), as well as the 'coherence' scale of the BIPQ ($p =$

		Follow-up at 3 months.		validated Symptom Checklist (SCL-90-R).		0.02, g = 1.70). Effect sizes provided are within group calculations.
Schaffer et al (2017)	33 children, 9-14 years. <1 seizure in past 12 months. Purposive sample. Israel.	Matched pairs design. Non-epilepsy control group. No follow-up.	Modular intervention with 2 five-week modules; memory skills training, and psychosocial training informed by CBT methods and techniques.	Youth self-report subtest (YSR), General Perceived Self-Efficiency scale (GSE), Children's Self-Control scale (CSC), and the Youth Life Orientation Test (YLOT). Parents completed the Child Behavior checklist parents form (CBCL).	ANOVA, Chi-square.	Between subject analysis showed modest intervention effect for optimism (YLOT) ($p < 0.05$) as well as self-efficacy (GSE) ($p < 0.05$).
Svanstrom et al. (2023)	15 children, 8-13 years. 2+ historical seizures, medication controlled. Convenience sample. Sweden.	One group, pre-test/post-test (before intervention and 3-month follow-up).	Psychoeducational intervention delivered by psychologists in groups (3-5 children). Mixed in-person and online delivery due to COVID-19 related restrictions.	ADHD-RS-IV Inattention subscale, the Behavior Rating Inventory of Executive Function, Second Edition (BRIEF2), Strengths and Difficulties Questionnaire (SDQ), DISABKIDS generic, and DISABKIDS epilepsy.	Paired sample t-tests.	Statistically significant reduction in self-identified executive function difficulties ($p = 0.03$, $d = 1.10$), as well as generic quality of life for self-report ($p = 0.043$, $d = 0.57$) and parent report ($p < 0.001$, $d = 1.40$) measures.
Tajrishi et al. (2015)	30 children, 14-18 years. No seizure in past 6 months. Medication controlled. Convenience sample. Iran.	Semi-experimental design with pre-test and post-test measures. Control group. Follow-up at six weeks.	Intervention group attended 11 sessions (2x per week, 45 minutes) and received an attribution retraining program, as well as communication training, anger management, and life skills training. Attribution retraining program based on Bandura, Seligman, and Wiener's models.	General Health Questionnaire (GHQ).	ANCOVA	Statistically significant reduction in mental health difficulties in all subscales of the GHQ; physical symptoms ($p = 0.01$, $g = 1.86$), anxiety and insomnia ($p = 0.01$, $g = 2.26$), social dysfunction ($p = 0.01$, $g = 2.03$), depression ($p = 0.01$, $g = 2.56$), as well as the overall index ($p = 0.01$, $g = 2.62$). Within-subjects effect sizes provided.

3.3 Sample Size and Characteristics

All studies required a diagnosis of epilepsy from a healthcare professional for participation. The characteristics of the sample did however vary in their presentation, with differences between studies with regard to their inclusion and exclusion criteria relating to time since last seizure.

Only three studies (Brown et al., 2019; Guven et al., 2020; Tajrishi et al., 2015) calculated power sizes to determine an appropriate sample size which informed participant recruitment. One study provided a justification for not using a power calculation to guide recruitment (Dorris et al., 2017).

There was variability in the overall sample sizes (including control conditions) for the studies. One study recruited a large sample of over 100 participants (Brown et al., 2019). Five studies had a sample size of less than or equal to 30.

It is also of note that inclusion and exclusion criteria were inconsistent between studies with regard to cognitive ability. For example, Svanstrom et al. (2023) applied an inclusion criterion of FSIQ ≥85 but based this on clinical judgement, whereas Schaffer et al. (2017) used a prediction of IQ (ESIQ) based on the completion of a WISC subtest (block design) and used a cut-off of ≤79 for exclusion.

Seven of the ten included studies included a control group, and five of these seven studies used randomisation (Brown et al., 2019; Dorris et al., 2017; Gurhopur et al., 2018; Guven et al., 2020; Tajrishi et al., 2015).

3.4 Effect Size

Effect sizes were provided for two of the ten included studies (Dorris et al., 2017; Svanstrom et al., 2023). For studies that provided means and standard deviations, however not an effect size, these were calculated for the main significant findings (Eom et al., 2016; Gurhopur et al., 2018; Guven et al., 2020; Rizou et al., 2017; Tajrishi et al., 2015).

Effect sizes were calculated using the formula: $$\mathbf{d/g=} \frac{\mathbf{M^1 - M^2}}{SD_{pooled}}$$

This was used for within subjects and between subjects comparisons. A recent analytic review highlights that this is the optimum calculation (Goulet-Pelletier & Cousineau, 2018).

Effect sizes were interpreted using Cohen's (2013) benchmarks of 0.2 (small), 0.5 (medium), and 0.8 (large). Where participant numbers were low (fewer than 30), 'Hedge's g' was favoured over 'Cohen's d' (Grissom & Kim, 2005). If effect sizes were provided within the study, these were not transposed based on sample size criteria.

3.5 High Quality Studies

Of the four high quality studies, three were RCT's (Brown et al., 2019; Dorris et al., 2017; Guven et al., 2020), whereas one utilised a matched pairs design (Schaffer et al., 2017). Only one high quality study (Schaffer et al., 2017) explicitly used a blinding process within their methodology, and one study provided a justification for not using blinding (Dorris et al., 2017).

Two of the studies carried out a power calculation before proceeding with recruitment (Brown et al., 2019; Guven et al., 2020) and another provided a justification for not conducting a power calculation to inform recruitment (Dorris et al., 2017).

None of the studies rated as high quality explicitly stated any potential harms that may have arisen from participation in the intervention, which is considered as part of the rating using the CCAT.

3.5.1 Intervention Outcomes

Two studies rated as high quality focussed their outcome measures on quality of life and the reduction of distress (i.e. anxiety, depression, emotional distress) (Brown et al., 2019; Dorris et al., 2017). One of these studies also measured changes in self-efficacy and epilepsy knowledge (Dorris et al., 2017).

Guven et al. (2020) had self-efficacy and epilepsy knowledge as intervention goals, as well as changing their participants attitudes towards illness. Schaffer et al.'s (2017) study focussed on self-efficacy and attitudes towards life (i.e. optimistic or pessimistic) as the

psychosocial aspects of their intervention. They also collected neuropsychological outcomes (memory-based, and executive function) as part of their study, commenting on the moderation effects that these may play.

All high-quality studies used validated and standardised psychosocial measures. These were used in addition to measures that were created for the purpose of the research. Brown et al. (2019) captured physical activity markers in their study, creating a tool to do this. Dorris et al. (2017) created participant and parent questionnaires for their study, which enabled the collection of qualitative and feasibility data.

3.5.2 Intervention Methods

Two of the studies used a modular CBT group therapy intervention using multiple sessions (Dorris et al., 2017; Schaffer et al., 2017). Schaffer et al. (2017) also included modular skills training for memory and executive function difficulties in their intervention.

Of the two studies that did not contain a primary CBT-based element, one (Brown et al., 2019) implemented a longitudinal exercise-based intervention, which included weekly or fortnightly sessions with the research team that utilised behaviour change techniques. There was also a psychoeducational aspect of this intervention. The final intervention (Guven et al., 2020) saw participants being given access to a web-based educational platform for 12 weeks, alongside reminders to use the platform and virtual technical support. There was no therapeutic intervention delivered by a clinician in this study, rather it was a remote psychoeducational intervention.

3.5.3 Intervention Effectiveness

Only one of the high-quality studies provided effect sizes for their main findings (Dorris et al., 2017). One further study provided means and standard deviations for the primary findings, which enabled calculation of effect sizes (Guven et al., 2020). The remaining two studies did not provide effect sizes or sufficient data to calculate these for their primary findings (Brown et al., 2019; Schaffer et al., 2017).

Of the studies for which effect sizes are calculated, Dorris et al. (2017) found a significant improvement in epilepsy knowledge after their CBT-based intervention which included

psychoeducational components (small effect size). This increased at three-month follow-up (medium effect size) suggesting that participants had continued to independently learn more about their epilepsy. There were no changes on measures of anxiety/depression likely reflecting the low baseline scores on these measures. The authors also reported very high acceptability and feasibility data including significant improvements in self-reported confidence in speaking to others about their epilepsy. Guven et al. (2020) reported significant results across all outcome measures for their web-based psychoeducational intervention. This included a significant increase in epilepsy knowledge (large effect size), seizure self-efficacy (large effect size), positive attitudes towards health (large effect size), as well as health literacy (large effect size).

Of the studies for which effect size was not calculated, Brown et al. (2019) did not report any significant results for their physical activity intervention. The research team discussed the high baseline scores for physical activity in the intervention group as a possible factor as to why changes did not reach significance. Schaffer et al. (2017) reported significant results for between subject analysis for their neuropsychological and CBT-based intervention for self-efficacy and optimism.

3.6 Acceptable Quality Studies

Of the four acceptable quality studies, one was a randomised controlled trial (Gurhopur et al., 2018), two used a quasi-experimental design (Rizou et al., 2017; Tajrishi et al., 2015) with non-randomised control groups, and one used a single system pre-test, post-test design (Svanstrom et al., 2023).

One acceptable quality study calculated power sizes to determine their sample size prior to data collection (Tajrishi et al., 2015), none of the other studies rated as acceptable did this (Gurhopur et al., 2018 Rizou et al., 2017; Svanstrom et al., 2023).

3.6.1 Intervention Outcomes

Out of the four acceptable quality studies, two were primarily concerned with the reduction of distress / mental ill-health (Rizou et al., 2017; Tajrishi et al., 2015), and two were primarily concerned with increasing quality of life (Gurhopur et al., 2018; Svanstrom et al., 2023). One study was also focussed on the reduction of executive

function difficulties, and this was reflected in their sampling inclusion criteria (Svanstrom et al., 2023).

All four studies used clinically valid and standardised instruments to measure the impact of their interventions. Three studies exclusively used self-report measures, two for children alone (Rizou et al., 2017; Tajrishi et al., 2015), and one (Gurhopur et al., 2018) used child self-report and parental-self report measures, as they included parental participants. One study (Svanstrom et al., 2023) used informant (parental plus teacher) report measures, in addition to self-report measures. The use of informant measures represents a methodological strength, reducing bias.

3.6.2 Intervention Methods

Three of the studies used a psycho-educational approach for intervention (Gurhopur et al., 2018; Rizou et al., 2017; Svanstrom et al., 2023). Rizou et al. (2017) furthered this approach by using a Socratic exploration of participant fears about epilepsy. One of these three studies carried out the intervention in one four-hour session after consultation with parents that this would be the most practical approach (Rizou et al., 2017), whereas the other two studies operated a modular design with multiple intervention sessions (Gurhopur et al., 2018; Svanstrom et al., 2023). One study also ran a parallel modular education program for parents alongside the children's programme (Gurhopur et al., 2018).

Tajrishi et al. (2015) used a skills training approach for their intervention, with a focus on attribution retraining. Limited information is provided about the development or adaption of this program, beyond that it had been previously used in the same country (Iran) for children with dyscalculia. The course was delivered across 11 sessions, each lasting 45 minutes.

3.6.3 Intervention Effectiveness

Only one of the acceptable quality studies (Svanstrom et al., 2023) reported effect sizes. The three other acceptable quality studies (Gurhopur et al., 2018; Rizou et al., 2017; Tajrishi et al., 2015) did however all provide means and standard deviations, which enabled the calculation of effect sizes. Gurhopur et al. (2018) found that their modular

education programme significantly increased children's epilepsy knowledge (large effect size), whilst also significantly increasing their seizure self-efficacy and quality of life, however the effect sizes for these were small. Rizou et al. (2017) found that their brief self-regulation intervention (one session, four hours) significantly reduced psychological distress and sleep problems (both large effect sizes). The very small sample size (n = 12 in the intervention condition) limits the generalisability of these findings. Svanstrom et al.'s (2023) psychoeducational intervention significantly reduced self-reported attentional difficulties (large effect size) and significantly increased self-reported quality of life (medium effect size). The parental (informant) reported quality of life also increased significantly (large effect size). Tajrishi et al.'s (2015) attribution retraining program led to a significant reduction in mental health difficulties (large effect size). The post-test in their study was conducted at six weeks, which provides some insight into longitudinal benefits of the intervention, however it is limited by the small sample size (n = 15 in the intervention condition).

3.7 Poor Quality Studies

Owing to methodological weaknesses, the two poor quality studies (Batista et al., 2015; Eom et al., 2016) in this review will be described in limited detail. Both studies utilised a single system research design (one group, pre-test and post-test), with no longer term follow-up. This reduces the validity and practical application of the findings, as no certainty can be drawn with regard to whether the intervention was the agent of change. These issues are exacerbated by low sample sizes of 17 (Batista et al., 2015) and 10 (Eom et al., 2016) respectively. No power analysis was carried out to determine an appropriate sample size in either study. Batista et al. (2015) created two outcome measures used in their study, which means that they were not validated or standardised, whereas Eom et al. (2016) exclusively used validated measures.

Batista et al. (2015) found that delivering manualised computer-assisted CBT increased children's epilepsy knowledge and stress knowledge. It also increased their frequency in using positive strategies for coping with stress. They did not, however, provide effect sizes for these and the lack of reported means and standard deviations meant that these could also not be calculated. Eom et al. (2016) found that a 35-week exercise programme significantly increased participants general health and quality of life. Effect sizes were not

provided in the study, however we were able to calculate them (g = 2.62 for general health; g = 2.47 for quality of life). These represent (very) large effect sizes, however the aforementioned methodological weaknesses impact the internal and external validity of both of these studies, and so these significant findings should be interpreted with caution.

4. Discussion

4.1 Evidence for Psychosocial Interventions

The present review demonstrates that the evidence base for psychosocial interventions for CYPE continues to grow and develop. The findings are promising with regard to the psychoeducational aspect of interventions increasing participants epilepsy knowledge, with four of the included studies reporting significant changes in this regard (Batista et al., 2015; Dorris et al., 2017; Gurhopur et al., 2018; Guven et al., 2020). The present review also highlights the role of psychosocial interventions in increasing the quality of life of CYPE, with significant findings found in three of the studies (Eom et al., 2016; Gurhopur et al., 2018; Svanstrom et al., 2023). Limited, missing, or non-significant longitudinal data within the included studies limits the extent to which the longevity of these changes can be assessed. Other significant outcomes included the reduction of distress (Guven et al., 2020; Tajrishi et al., 2015) as well as increased self-efficacy and problem-solving skills (Batista et al., 2015; Guven et al., 2020; Schaffer et al., 2017), and improved confidence in talking about epilepsy with others (Dorris et al 2017).

The robustness of the evidence base is impacted by a tendency for participants to be recruited through convenience or purposive sampling as a result of receiving hospital care (100% of studies included in the present review). This adds bias into the sample, and it is important that there is transparency around how samples are selected from treatment databases, particularly with a purposive sample. In addition, when interventions are delivered as part of clinical practice within a treatment setting it is often impractical or impossible to blind participants (e.g. if they are part of a waiting list control group), which limits the internal validity of the research.

4.2 Treatment Components

The majority of studies included in the present review utilised evidence-based treatment components, which are supported by the literature. The most common component within the interventions was psychoeducation (9/10 studies), which meets the evidence-based practice suggestion from Michaelis et al. (2018a) that "each patient with epilepsy should receive psychoeducation".

With regard to affective challenges for people with epilepsy (e.g. depression and anxiety), the studies included in the present review utilised behavioural and skill-based approaches in addition to interventions informed by CBT and mindfulness practices. This aligns with the evidence-based practice suggestion from Michaelis et al. (2018a) that "treatment components may include behavioural intervention (e.g. social activation) and skill-based interventions (e.g. problem solving, social skills training)" when working with depression, and with regard to anxiety "the highest level of evidence pertains to the implementation of mindfulness exercises." Six of the ten studies included in the present reviewincluded a direct skill-training element, which could include mindfulness-based exercises.

Cognitive behavioural therapy continues to be a leading evidence-based intervention for people with epilepsy, with a recent systematic review and meta-analysis suggesting CBT-based interventions have led to better outcomes with regard to depression and quality of life (Li et al., 2023). Four of the ten studies included in the present review had an explicitly stated CBT-based approach to their intervention. Interestingly, the same authors (Li et al., 2023) found that CBT delivered in an individual capacity had a larger effect size than group-based CBT. In the present review, CBT-based interventions were delivered in group settings, including in the two studies rated as high quality (Dorris et al., 2017; Schaffer et al., 2017).

4.3 Intervention Goals and Outcome Measures

Studies included interventions that aimed to increase epilepsy knowledge, increase quality of life, reduce distress, and develop skills and self-efficacy. Across the ten included studies, these intervention goals were operationalised through diverse interventions limiting the generalisability of findings and clinical applicability.

All ten of the included studies utilised standardised outcome measures for a substantial part of their data collection and analysis. Only one study relied on bespoke measures for most of their outcome data (Batista et al., 2015). This represents a positive change since the earlier review (Corrigan et al., 2016). Outcome measures were however heterogenous, which presents a challenge when comparing results between studies and developing a coherent and consistent evidence base. For example, in this review we saw three different quality of life outcome measures, each using different items and constructs. Adopting a consensus on outcome measures between research groups would support higher quality research, as well as the ongoing standardisation of those measures. In their systematic review of quality of life instruments for children with epilepsy, Crudgington et al. (2020) recommend the use of the Quality of Life in Childhood Epilepsy Questionnaire (QoLCE-55) and the Health-Related Quality of Life Measure for Children with Epilepsy (CHEQoL). Of these, the CHEQoL is the only one that provides a child self-reported health-related quality of life score, and so perhaps should be favoured. Cultural differences in the concept of quality of life may however limit global adoption of a single measure.

Of the ten included studies, only two reported longitudinal outcome data beyond 4 months (Brown et al., 2019; Dorris et al., 2017), four studies had no follow-up data, and four studies did not have a follow-up after 4 months. Dorris et al. (2017) showed sustained treatment effects at 6-month follow-up in relation to increased epilepsy knowledge and in confidence talking about epilepsy. Longer term follow-up periods should perhaps be a greater methodological consideration when constructing interventions and research studies focussed on psychosocial interventions for CYPE, given the evidence base for accelerated forgetting in this population more broadly (see Butler & Zeman, 2008 for a review). One of the included studies combined a memory training program with their psychosocial intervention (Schaffer et al., 2017), however there was no longer-term follow-up.

4.4 Future Research

The included studies varied significantly between their inclusion and exclusion criteria, such as the extent to which participants were seizure free or experiencing active seizures. For example, Brown et al. (2019) and Rizou et al. (2017) required participants to have

experienced a seizure in the past 12 months, whereas other studies required differing periods of seizure-free status prior to participating, or for epilepsy to be medically controlled. One study actively included participants with neurodevelopmental difficulties (relating to attention), whereas other studies specifically excluded CYPE who had neurodevelopmental co-morbidities. A solution to this could be to encourage agreement between researchers for a collective set of parameters, which could be achieved through large-scale multi-centre research collaborations.

When the collective sample between studies does not combine into a homogenous group, the rationale for excluding people with cognitive impairment could be called into question. For example, Svanstrom et al. (2023) stated their exclusion criteria for 8–11-year-olds of an IQ <85 was a "pragmatic decision based on the clinical judgment that they were likely to have difficulties accessing the content of the intervention due to cognitive and reading ability". Such views have been challenged within the adult population by reviews of the evidence base, such as Vereenooghe & Langdon (2013), who found a moderate effect size for psychological therapies for people with intellectual disabilities. Although excluded from the present review due to the sample containing participants with a learning disability (legacy exclusion criteria), we scored one promising study using the CCAT and found that it would have been included as a high-quality study (Appendix B). Bennett et al. (2021) found significant improvements in self-reported mental health problems, the impact of mental health problems, anxiety and depression symptoms, as well as quality of life (all medium effect size) for CYPE. Their sample contained 9/23 children with a learning disability (39.1%). Although the sample size was small, this tentatively suggests that children and young people with a learning disability may positively respond to a phone-based CBT intervention, and reduces the rationale for their exclusion in other studies. This is a point of consideration, should this review be updated in the future, as well as for the research area as a whole.

As previously stated, the standardisation of outcome measures across studies would also strengthen the evidence base. If there could be agreed guidelines for which outcome measures to use, then this would support good practice. There are existing systematic reviews that can guide this process (e.g. Crudgington et al., 2020), however this needs to be developed for all outcome measures.

4.5 Limitations and Strengths

There are a number of limitations of the present review. The review is subject to publication bias, in that studies with significant findings are more likely to be published and therefore included in this review. Only one included study did not report any significant results (Brown et al., 2019). The heterogeneity of the included studies also presents a limitation, as the differences in populations, interventions, and outcome measures mean limits the extent to which pooling the results leads to a coherent picture of the research landscape. This heterogeneity also restricted our ability to perform a meta-analysis, which represents a weakness of the present review. In addition, whilst the proportion of high-quality studies (40.0%) has increased since the earlier review (17.6% in Corrigan et al., 2016), 60% of the included studies were either of poor or acceptable quality and as such there are notable methodological weaknesses that negatively impact their internal and external validity.

Strengths of the present review include the use of the CCAT, which enables a quality appraisal of studies with a variety of methodologies and was developed to address the dearth of consistent reliability and validity data among critical appraisal tools (Crowe & Sheppard, 2011). The CCAT is also accompanied by user guidance which adds to its validity through increasing the uniformity of its application. Rating pairs were also used in the present review at two stages; title and abstract searching, and during quality rating. This strengthens the present review through the reduction of subjectivity bias, errors, and increasing transparency.

4.6 Summary and Conclusions

There is an expanding body of research that underscores the efficacy of psychological interventions for children and adolescents diagnosed with epilepsy. This body of evidence is steadily improving in terms of both quantity and quality. Effective elements within these interventions include psychoeducation, strategies grounded in cognitive-behavioural therapy principles, and mindfulness techniques. This aligns with the evidence-based guidelines established for adult populations. Treatment goals for these interventions focus on enhancing the overall quality of life, mitigating symptom-related distress, and bolstering knowledge and skills. The instruments used to measure these

outcomes are predominantly standardised, however remain heterogeneous between studies, which introduces a degree of variability that may affect the overall strength of the evidence base.

Since the previous review there have been a number of studies employing an RCT design, which represents progression for research into the efficacy of psychosocial interventions for CYPE. The proportion of studies that have been rated as high quality has also increased significantly since the previous review, so too has the proportion of studies using standardised outcome measures. The evidence base continues to be limited by the heterogeneity of the samples, reliance on convenience and purposive sampling, as well as significant variability in the outcome measures used. The earlier review concluded that "the adoption of multi-centre collaborations may overcome many of the methodological limitations observed in the current evidence base" (Corrigan et al., 2016), which remains true today.

4.7 Declaration of Interests

The authors declare that they have no known competing financial or personal interests.

4.8 Other Information

This systematic review is registered on PROSPERO, reference: CRD42023424140.

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Chapter 2

What is the Process of Coping and Adjustment when Parenting a Child with Dravet Syndrome? A Qualitative Exploration

Prepared in accordance with the author requirements for 'Epilepsy and Behaviour'

Link (Full 2023 Author Guidelines): https://osf.io/bwnr2

Plain Language Summary

Background

Dravet syndrome is a serious condition which can lead to severe seizures as well as learning difficulties. Parents of children with Dravet syndrome are often required to provide a lot of care for their children, which can have a huge impact on their quality of life. We aimed to try and understand how parents cope and adjust to the demands of caring for a child with Dravet syndrome.

Questions Addressed by this Study

1. What is the process of coping and adjustment when parenting a child with Dravet Syndrome?

How this was Done

This was done by running five focus groups with parents of children with Dravet syndrome. In these focus groups, we asked parents questions relating to their experience with regard to Dravet syndrome. We then wrote down what the parents said, which became our 'data'. This data was then analysed to create a theory for how parents cope and adjust to caring for a child with Dravet syndrome.

Results and Conclusions

We created a model to help understand the process of coping and adjusting to caring for a child with Dravet syndrome. Using this model, we suggest ways in which parents could be supported through this process.

Abstract

Purpose

Dravet Syndrome (DS) is a severe form of epilepsy that can require a large amount of caregiver input across the lifespan. This demand often falls on parents, who are faced with considerable challenges including physical demands, financial demands, and coping with pressures on mental wellbeing and increased risks of mental health difficulties. Given this considerable challenge, we believe that it is important to explore the ways in which parents cope and adjust to caring for a child/children with Dravet Syndrome (CwDS), and then consider how to effectively support these parents.

Aims

We aimed to explore the process of coping and adjustment that occurs for parents caring for a child who has a diagnosis of Dravet Syndrome.

Methods

Using a Grounded Theory methodology, we conducted five focus groups, each with 4-6 participants. These were run in Glasgow, London, Chesterfield and Manchester in partnership with Dravet Syndrome UK, a leading charity offering parent/carer support.

Transcripts of the focus groups were then recorded and coded into themes to generate a theory that is grounded in the data.

Results

We created a grounded theory of coping and adjustment with regard to parenting a child with DS. This included external factors (loss and insufficient resource) as well as an internal model of coping and adjustment, which encompassed a prominent theme of trauma and the ways in which parents respond to this trauma.

Conclusions

The study provides important insight into the ways in which parents cope and adjust to caring for a child with DS. Our theoretical model suggests further research into specific targeted therapeutic input for parents of CwDS. Therapeutic interventions should address the areas that negatively impact coping and adjustment, such as a sense of loss and guilt, as well as direct trauma work.

Keywords

Dravet Syndrome, Parental Coping, Caregiver Burden, Trauma, Loss, Epilepsy

1. Introduction

Dravet syndrome (DS) is a severe form of epilepsy, with a life-long prognosis, as well as life-limiting and life-threatening implications (Dravet, 2011). The incidence of DS is rare, with a previous estimate of UK prevalence as around 1:40,900 (Brunklaus et al., 2012), however the burden of care experienced by those who look after children with DS is severe, given the associated cognitive impairments, behavioural challenges, and physical disability (Brunklaus et al., 2012), as well as the risk of sudden unexpected death in epilepsy (SUDEP) (Dravet, 2011).

Nolan et al. (2006) identify three stages of condition progression for DS, from the initial identification and diagnosis through to maturity (adult life). These stages bring with them different demands on caregivers in terms of functional roles, for example from temperature monitoring to finding suitable education and long-term care (Camfield et al., 2012; 2016), but there are also significant demands in terms of coping and adjustment to these challenges. Nolan et al. (2006) conducted semi-structured interviews with parents exploring their circumstances, which they summarised as follows:

> *"Parental experiences evolve from terrible anxiety about the diagnosis to extreme stress over constant seizures…eventually they become resigned to a life with restricted social contact but find more personal contentment."*
>
> Nolan et al. (2006)

This language removes a sense of agency from parents, and perhaps over-simplifies the collective experience. There is a dearth of literature that qualitatively explores the experience of that journey from a parent's perspective, and the individual routes that people take with regard to coping and adjustment. In their review, Jensen et al. (2017) recognise this notable absence in research, whilst also identifying "the impact on relationships with friends, family, and the spouse (and related social isolation), sleep problems, financial stress, work, …grief and general emotional stress among caregivers" (Jensen et al., 2017).

Churchill et al. (2010) found that parents (of children with significant disabilities) who have developed better coping strategies, report fewer depressive symptoms – highlighting an intuitive yet important link between the two. The ways in which parents

develop and apply their skills in adjusting to the circumstances that befall them is less clear, with greater focus paid to the presence of significant events, rather than how parents internalise those events and adjust. For example, Nabbout et al. (2018) found five factors that have the greatest impact on the experience of DS for the family, these were:

- seizures
- expressive communication of the child
- receptive communication of the child
- impact on daily activities
- social functioning of the caregiver

Previous, survey-based, research has explored practical implications arising as a result of the burden of care. Frequently one or both parents reduce their working hours, which necessarily causes financial difficulties (e.g. Campbell et al., 2018; Jensen et al., 2017). This is not the only way in which families are financially disadvantaged, as they contend with the cost of medication, emergency transportation, and diagnostic procedures, as well as numerous other costs (Jensen et al., 2017; Strzelczyk et al., 2014).

Campbell et al. (2018) found that managing behavioural problems was the greatest difficulty for parents, and the greatest (measurable) impact on them was anxiety and / or depression (70% reported more than 'slight problems' and 34% more than moderate problems). In their review of the literature, Gonçalves et al. (2021) identify that the mental health and quality-of-life of informal carers (e.g. parents) are compromised, however that very few studies report factors associated with increases in depression and anxiety.

A family's interaction with medical services can also support positive adjustment; such as the potential for genetic diagnosis to facilitate support (Brunklaus et al., 2013). Diagnosis may however represent a negative experience (Goodwin et al., 2015), suggesting individual differences between experiences. Interaction with medical services can also be mediated by other factors, such as socioeconomic status (SES), with Jensen et al. (2017) finding that families with lower SES accessed professionals through emergency department visits and hospitalisations more often, rather than pre-planned visits to neurologists. This may necessarily impact the experience of the CwDS, as well as their family – potentially impacting the adjustment process.

Whilst there is sparse psychosocial research directly linked to DS (Jensen et al., 2017), parental adjustment processes have been explored with other diagnoses which whilst not being directly comparable, may prove insightful. Grootenhuis & Last (1997) outline the critical role of hope with regard to parental mental health during serious child illness. The authors however also highlight the negative implications of parents having 'illusory control' – suggesting that there is a sweet spot between optimism and illusion for parental coping. Muscara et al. (2018) propose three coping trajectories for parents following a serious injury or illness for their child. These were 'resilient', 'recovery', and 'chronic'. Differences in these trajectories were due to parental psychological factors (e.g. mood and anxiety), rather than demographic factors or illness characteristics.

The demands of parenting a CwDS means that caregivers are required to become 'medical professional parents' (Camfield et al., 2016), in addition to numerous other roles (e.g. carer). When combined with loss of employment (Campbell et al., 2018), and the identity that this provides, it can be challenging for parents to retain a sense of independent identity, which may further impact their adjustment. The National Institute for Clinical Excellence guidelines (NICE, 2020) recognises the importance for carers to have space away from those that they care for to discuss their needs, which can support the identification of their role, the demands of that role, and the consideration of appropriate support.

1.1 Research Aim

The present study aims to take an inductive approach to understanding the parental coping and adjustment that occurs when parenting CwDS, with the hope that this may play a formative role in identifying possible future key areas for the provision of supportive interventions.

1.2 Research Questions

1.2.1 Primary Question

1. What is the process of coping and adjustment when parenting a child with Dravet Syndrome?

1.2.3 Secondary Questions

- What are the experiences of parent/carers of children with Dravet Syndrome?
- What adjustments are required in caring for a child with Dravet Syndrome?
- What influences resilience and coping over time when caring for a child with Dravet Syndrome?

2. Methodology

An inductive qualitative approach was utilised to form an initial theoretical understanding of the ways in which parents cope and adjust to caring for a child with DS. Grounded theory was used to construct a working model as the basis for future research. A constructivist grounded theory approach was selected as it enabled the exploration of parental experience over time and how this had been processed. It also allowed for consideration of the inherent individuality of participants, recognising that the focus groups were a specific snapshot of time, and a product of all the people who were involved. We were also keen that the analysis created a working model, extending beyond the remit of thematic analysis.

The primary researcher for the present study was Anthony Mercier, and the secondary researcher was Professor Liam Dorris.

2.1 Ethical Approval

Ethical approval was granted by the University of Glasgow College of Medicine, Veterinary & Life Sciences (MVLS) committee on 22/09/2022 (Appendix D).

2.2 Participants

Participants were recruited to the study between October 2022 and February 2023 through communications from Dravet Syndrome UK (DSUK), who are a UK-based charity working with families directly impacted by DS. As such, participants were recruited from a convenience sample.

Communications were sent to prospective participants via email mailouts to members of the charity, as well as through closed-group messages on social media. Prospective participants were then invited to take part in focus groups.

Prospective participants were then provided with the following documentation to read and complete, and were given an opportunity to ask any questions before signing up for participation:

- Participation information sheet (Appendix F)
- Privacy notice (Appendix G)
- Demographic survey (anonymous) (Appendix H)
- Participant consent form (anonymous) (Appendix I)

Inclusion criteria for participants:

- Parent or legal guardian of a child with Dravet syndrome
- Aged 18 years or older
- Member of DSUK, which requires medical diagnosis
- English speaking (fluent)
- Able to participate in a focus group lasting up to 120 minutes

Exclusion criteria for participants:

- Have great difficulty hearing, reading, or communicating that would be prohibitive to the chosen methodology
- Have severe neurological or cognitive deficits or a psychiatric condition that is not managed (e.g. through medication) and that may affect their ability to participate in a focus group

2.3 Materials

A question schedule was developed (Appendix J) to guide the focus groups. These questions were initially developed by the primary researcher and then modified through discussion with a panel of professionals and people with lived experience of Dravet syndrome. This informed how the questions were phrased to anchor participants in the early experience of Dravet (e.g. 'time of the first seizures'), as well as to provide guidance on the suitability of questions and number of questions to be asked.

Focus groups were recorded using a highly secure encrypted (256-bit AES encryption) and password protected Dictaphone and two boundary microphones.

2.4 Procedure

Participants were invited to attend one in-person focus group in either Glasgow, London, Manchester, or Chesterfield, lasting 90-110 minutes. The primary and secondary researcher co-facilitated the first focus group, with the primary researcher then facilitating focus groups two to five.

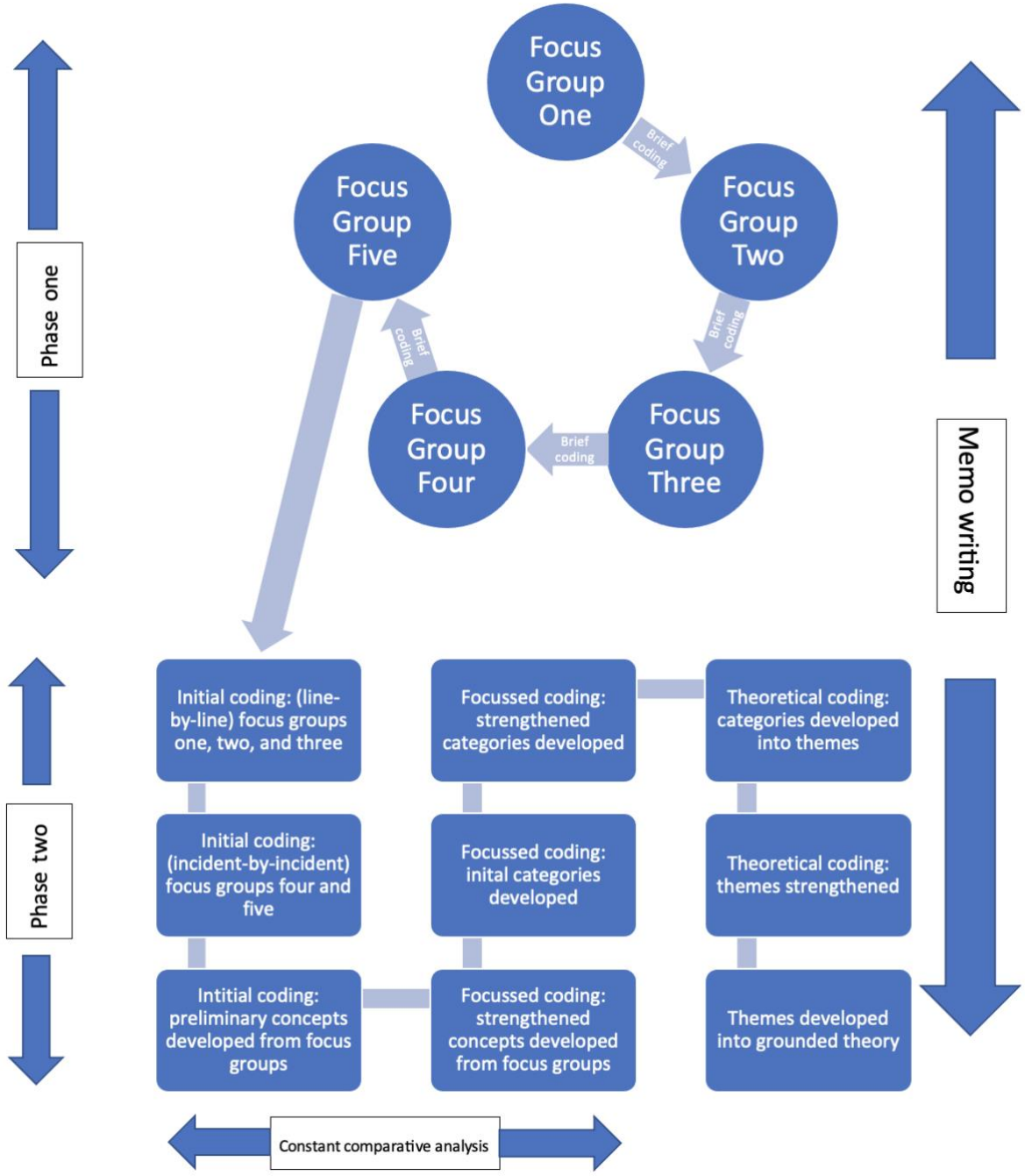
Audio data was manually transcribed by the primary researcher within one week of the focus group being completed, and the audio file was deleted. Any identifiable information was removed from the written data.

A session was then held with a panel of DSUK staff and research participants to present the themes that had arisen from the data and to listen to their reflections on these and how they were presented.

2.5 Data Analysis

Figure 2.1

Data Analysis Process



2.5.1 Phase One

Brief coding carried out between the focus groups supported 'theoretical sampling', which has been described as "identifying and pursuing clues that arise during analysis in a grounded theory study" (Birks & Mills, 2015 p68). This facilitated identification of theoretical strands to address with follow-up questions during subsequent focus groups, which enabled simultaneous data collection and analysis, which is a core facet of Grounded Theory (Charmaz, 2006).

> *Interviewer: "You talked, and people have phrased it in different ways, but about always being alert. Always watching…"*
>
> *(Focus group five)*

2.5.2 Phase Two

Constant comparative analysis was carried out during initial coding and focussed coding. The developed codes were compared within focus groups and between focus groups to construct a grounded theory. Initial coding followed Charmaz's (2006, p48) guidance to "look closely at actions and, to the degree possible, code data as actions".

Charmaz (2006, p48) states: "initial codes are provisional, comparative, and grounded in the data" and "speed and spontaneity help in initial coding." Initial coding is a creative and draft process to be revised through constant comparative analysis. We conducted line-by-line coding on the first three focus groups, and incident-by-incident coding on groups four and five. All data was then reviewed during focussed coding. Charmaz (2006, p57) defines focussed coding as "using the most significant and/or frequent earlier codes to sift through large amounts of data." Focussed coding was carried out until theoretical categories were saturated.

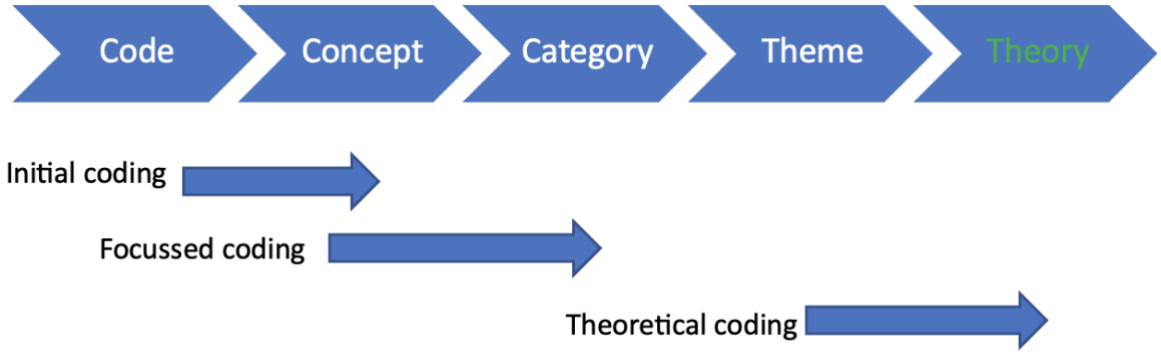
Theoretical coding was then applied to the focussed codes to generate a grounded theory. Glaser (1978, p78) described this final step as "how the substantive codes may relate to each other as hypotheses to be integrated into a theory." This has been adopted by

Charmaz (2006) in her Constructivist Grounded Theory. During this phase Qureshi & Ünlü (2020) describe two main processes: theoretical sorting and generating hypotheses. During this stage, no new data is analysed.

During phase two of the analysis, the Ünlü-Qureshi instrument (Qureshi & Ünlü, 2020) structured the development of a grounded theory. This four-step process is outlined in Figure 2.2. A fifth step is then the creation of a theory.

Figure 2.2

Adapted Diagram of Qureshi & Ünlü (2020) Instrument



2.5.3 Memo Writing

Memos were kept throughout data collection and analysis to support the creation of a grounded theory.

2.5.4 Constant Comparative Analysis

The simultaneous collection of data and analysis, as well as the revisiting of old data when new themes emerged ensured constant comparative analysis (Figure 2.1).

2.5.5 Coding Pairs

For data triangulation, two coding pairs were utilised. In the first coding pair, the primary researcher coded all of the focus groups, whilst the second coder provided initial coding

for 10% of each focus group. Disagreements were resolved through discussion. The second coding pair discussed the generated themes and how they combined into the grounded theory.

3. Results

3.1 Demographic Data

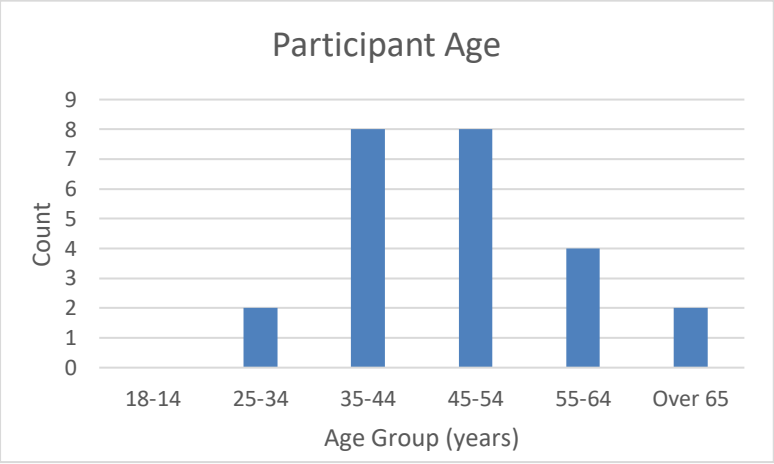
In total, 24 participants attended 5 focus groups.

22/24 participants identified as female, representing 92% of the sample. The remaining 2 participants identified as male (8%).

Two thirds of the participants (67%) were aged between 35 and 54 years (Figure 2.3). 14/24 (58%) participants' children had been diagnosed with Dravet syndrome more than 11 years ago, with 5/24 (21%) diagnoses between 0 and 3 years ago, and 5/24 (21%) diagnosed between 4 and 7 years ago.

Figure 2.3

Participant Age Ranges



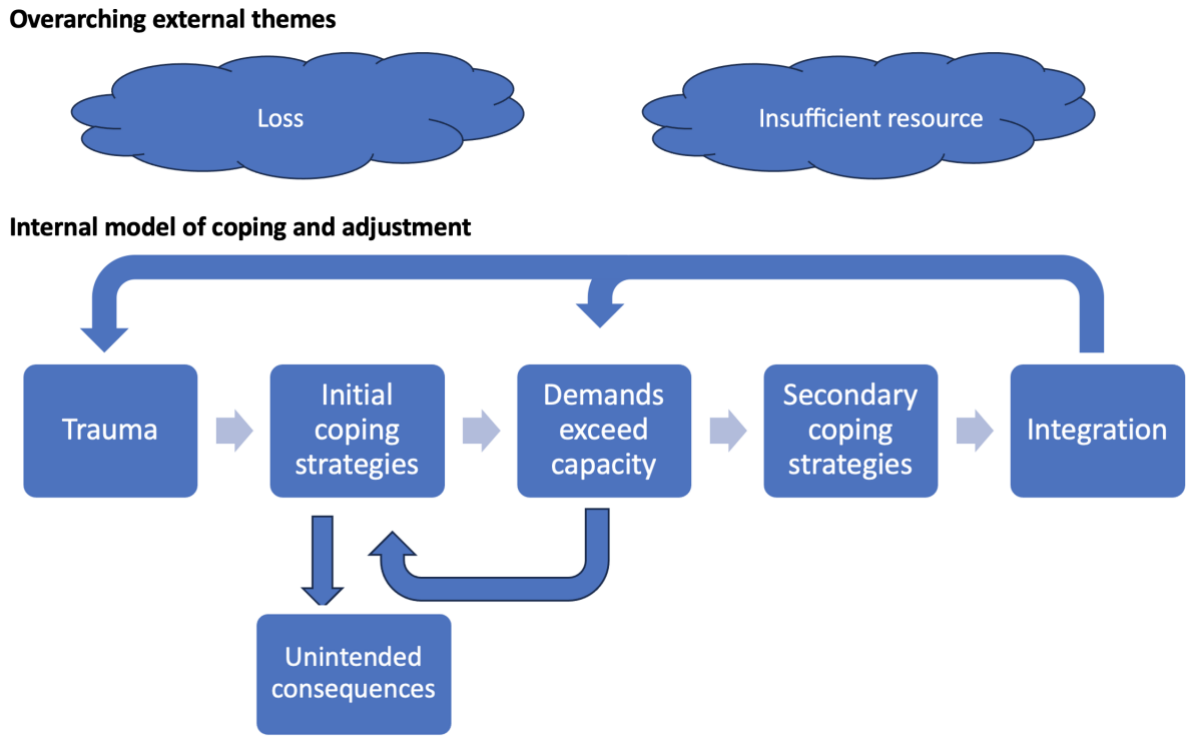
3.2 Grounded Theory

Figure 2.4 outlines a grounded theory from the data for how parents cope and adjust to caring for a child diagnosed with Dravet syndrome. The collected data was used to generate "overarching external themes" as well as an "internal model of coping and adjustment", which when combined constitutes the grounded theory.

A narrative of this, including the superordinate themes and categories, will accompany direct quotes from the focus groups (Tables 1 and 2), provided in brackets.

Figure 2.4

Grounded Theory Model of Coping and Adjustment



3.2.1 Overarching External Themes

Two overarching external themes provide the wider context for the internal model, and the challenges associated with coping and adjusting to parenting a CwDS.

Loss

Throughout the experience of coping and adjustment was a profound sense of loss, which could return poignantly at any time. The theme of loss captured a host of categories (Table 2.1).

One aspect was social loss, termed as 'isolation'. Parents found that people who were very close to them before withdrew (1a), or didn't know how to support (1b), or what to say (1c, 1d). Throughout all focus groups, parents reported shrinking social circles of family and friends (1e, 1f). Parents also described distancing themselves from others, as there became little overlap between their collective experiences (1g). This growing gap meant that communication between those that are Dravet parents and those who are not can become more difficult (1h). Bridging this gap in understanding is made infinitely more difficult when contending with the exhaustion associated with being a Dravet parent.

Other aspects of this theme related to a loss of what life could have been. Categories within this included "loss of 'normal' child", which was experienced deeply (1i), and was recurrent (1j). Parents also experienced "loss of a normal life" (1k), through having to stop 'normal life' activities, due to the demands of caring for a CwDS and insufficient resources or support, which in-turn causes changes in self-identity (1l). These feelings of loss return when parents encountered people, events, or objects that drew attention to the gap between their life and a previous notion of what their life 'should' be (1m), as well as in response to the behaviours of the child whom they care for (1n). Considerable guilt was associated with these losses, as parents may feel a responsibility for their child's condition (1o). This was reinforced through genetic testing (1p), and was carried by parents throughout their Dravet journey, extending to feeling guilty about what their other children may have lost too (1q).

Insufficient Resource

The second overarching theme centres around the insufficient resource available to parents. Key categories in this theme were "failed by healthcare" and "failed by social care" which were throughout the Dravet journey. Parents were more likely to feel failed by healthcare in the earlier parts of this journey (1r, 1s), which would understandably act as a precursor to some of the initial coping strategies (e.g. becoming a medical

professional parent (2t)), as well as the unintended consequences of that (e.g. becoming the only person who can provide care (2w)).

Parents feeling failed by social care typically happened later in the journey, for example when neurodevelopmental issues start to appear (1t), or when behavioural issues are more pronounced (1u). There was recognition that systems, however flawed, were in place for the child, however the same could not be said for the parents, who are left to fend for themselves, literally, in the absence of any support systems (1v).

These experiences necessitated parents to perpetually assume the roles of carer and advocate. Against a system that often felt like it does not value or care for them or their child, parents are left to do this for themselves (1w). One example role was that of an "eternal advocate" (1x, 1y), which required parents to build up their own medical knowledge, knowledge of health and social care systems, and to be forthright in their approach. Different rules and regulations associated with child and adult services mean that parents are continually required to update their knowledge (1x).

Table 2.1

Quote Table for Overarching External Themes

Ref.	Theme	Focussed Category	Quote
1a	Loss	Isolation	"I personally had no support – my family just didn't want to know"
1b	Loss	Isolation	"My family was too nervous, my mum was too nervous, and you do feel completely isolated."
1c	Loss	Isolation	A: "People tend, people do avoid you" B: "Absolutely" A: "They don't know what to say" B: "Absolutely"
1d	Loss	Isolation	"My best friend and my bridesmaid and the last message I sent her was (CwDS)'s diagnosis, and I haven't heard from her since."
1e	Loss	Isolation	"I have lost hundreds of friends on my Dravet journey"
1f	Loss	Isolation	"We are so used to…isolation, our circle of friends disappeared."
1g	Loss	Isolation	"Your world becomes so far removed…"
1h	Loss	Isolation	"And you feel like saying, excuse me – stop! Come and help us! And people say, they do say they understand – but they don't."
1i	Loss	Loss of 'normal' child	A: "That day your healthy baby died" B: "That is exactly what happened"
1j	Loss	Loss of 'normal' child	"You know when it hit me… was his fourth Christmas, and still being in the same aisle four years later in the toy shop… I have not progressed."
1k	Loss	Loss of 'normal' life	"It was never the same again. Never, no matter how much I tried for it to be the same. I tried to normalise things as much as I could. It just wasn't the same, it couldn't be."
1l	Loss	Loss of 'normal' life	"I was in a career and I was thinking I was going to go back… I suppose even now – even when I think I can, something always seems happen… I always see myself now as having lost confidence over the years, skills and abilities – and it is confidence."
1m		Loss of how life should have been	A: "…we haven't really talked about the grieving process…and I still think I go through that now…" B: "…it never goes" A: "…it never goes, and you will always you know I can remember not so long ago erm a friend of mines daughter who is the same age as (CwDS) had a first child" B: "Yeah, like the family line is over now, like…we were talking like maybe six or seven kids"
1n	Loss	Loss of 'normal' child	A: "I can think the last time I felt upset and angry – and erm (CwDS) had had her period, and she had left… took the sanitary towel out… opened her to put her socks in the drawer and she just shoved them in the… and you think…(sigh) (laughing in the group) B: "And I'm laughing because I get what you mean" A: "And I thought I ain't signed up for this…who has to put up with this?"
1o	Loss	Guilt	"My son died when he was (age)…he had it too… he died of a seizure, and myself, (CwDS), and (CwDS) have all got the SCN1A gene – so the guilt is there."

1p	Loss	Guilt	"I had passed the gene onto (CwDS) and so I have lived with that since he was tiny. So, I think that is why I am always in such a weird state with it because… there is a bit of that like 'god, I gave him that'"
1q	Loss	Guilt	"They become little carers, and you feel a lot of guilt, a lot of guilt don't you I think, but, because you know they shouldn't be doing that."
1r	Insufficient resource	Failed by healthcare	A: "So how did you manage in those few months before you were referred? Because you were dealing with seizures then and nobody was helping you…" B: "No, they couldn't do nothing, they said you have to wait for the doctor to call, the neurologist…it was really hard. That time I used to cry my eyes out, because I couldn't do nothing for him"
1s	Insufficient resource	Failed by healthcare	"I remember like about six doctors around his cot one time, and he started having a seizure, and I was like is someone going to give his rectal meds? Is anybody going to give him it or do I do it? And there was no answer from anybody…there was no help, there was no support, it was just left to you"
1t	Insufficient resource	Failed by social care	"…it took seven years to get a diagnosis of autism, and the first one that we went to was a shit paper with twenty questions, he didn't even look at (CwDS), he just talked to me and then said 'I think you are just chasing another title."
1u	Insufficient resource	Failed by social care	"There are some meetings that are an hour long for us… which are a waste of time, complete wate of time. I think he likes to just come in and have a sit down"
1v	Insufficient resource	Failed by social care	"I have been bitten, punched, kicked, headbutted, you know – bruises – pinched – all those things. And if I was doing that to her, she would be taken off me… but nobody looks at that – the impact it has on you."
1w	Insufficient resource	Failed by social care	A: "You don't get a package, you don't get someone saying ring this number, or ring that number…" B: "It is through word of mouth that you find things out… and you are fighting for every little thing. And you get to that stage when you think, I can't carry on… I can't carry on no more."
1x	Insufficient resource	Eternal advocate	A: "And your child mustn't grow up and become and adult, because that it a whole other thing." B: "…I mean this week I've had three-hour annual nursing assessment meeting, I had two-hour EHCP thing yesterday, it is just constant battles and fights, and paperwork" C: "I can't say that bit gets better, it probably does… and then you get to transition" A: "You get to know the words that you need to write on the forms…"
1y	Insufficient resource	Eternal advocate	"You end up basically being an advocate for your child, because they can't…"

3.2.2 Internal Model of Coping and Adjustment

The internal model relates to the personal process of coping and adjustment within the external context of repeated loss and continued insufficient resource.

Trauma

The theme of trauma was persistent and prominent. Parents provided examples of clear single-event trauma (2a, 2b), relating to situations when they believed their child was in mortal danger, as well as the looming spectre of SUDEP (2c). There were also examples of secondary trauma relating to medical procedures (2d), receiving news from medical professionals (2e), and receiving the diagnosis (2f). Arguments could be made, however, that given the profound impact on the parents' lives, these examples are direct trauma.

Traumatic memories created visceral re-experiencing when discussed (2g), were experienced in a dissociative way (2h), or were too painful to revisit (2i, 2j). Some participants referred to trauma specifically (2j), whereas others were not sure about the terminology, and perhaps awareness grew through discussion (2k).

In addition to specific events, the cumulative impact of repeated trauma (2l) meant parents lived in a heightened state for prolonged periods (2m). Parents referred to the continued rush of adrenaline during prolonged traumatic experiences (2n), and how this impedes their processing of the events (2o).

Initial Coping Strategies

Continued existence in "survival mode" alongside repeated traumatic experiences understandably led to the development of initial coping strategies (2p) borne through immediate necessity, without the space, time, and resource to consider the longer-term picture.

The most prominent initial coping strategies centred around "hypervigilance", with a combination of being in a prolonged heightened state, and feeling pressurised to spot everything with regard to DS (2q). This state of hypervigilance becomes the default state for parents (2r), with the strategy being used more and more. This cycle is reinforced

both by the times when seizures are caught ('I'm glad I was there') as well as the times when anything is missed ('I wish I had been there').

Parents described intense researching to find out as much about DS as they could (2s). This could be the formative steps to becoming a 'medical professional parent'. This search for information became all-consuming (2t, 2u). Conversely, a small sample of parents described an avoidant initial coping strategy (2v).

The unintended consequences of these initial coping strategies were felt in the short-term and the longer-term. Parents quickly develop such an extensive knowledge of their child and DS ("medical professional parent"), that they believe nobody can match their ability to care for their child (2w). When this combines with the reticence and anxiety of those who may have been able to support the parents (1b), finding support or respite becomes increasingly difficult.

These initial coping strategies are developed through necessity, and have positive aspects (2x), as otherwise they wouldn't be retained. The reinforcement from seeking and finding useful information, or catching a seizure event, encourages further use of that strategy. Equally, when there is a gap in knowledge, or events are not caught, the guilt associated with this served to drive hypervigilance and information seeking (2y). Indeed, a sustained sense of guilt acts as a powerful motivator for initial coping strategy use (2z). This continues beyond a decline in parental health (3a).

<u>Demand Exceeds Capacity</u>

Often, the demands of parenting a CwDS far exceeded a parent's capacity (3b), and the psychological impact of this could be extensive (3c). Unsustainable initial coping strategies, continued heightened state of vigilance, and inability to rest (2r) contributed to poor mental and physical health (3a). As well as ongoing exhaustion, there were specific times when this was felt acutely (3d). The cumulative effect of ongoing exhaustion and repeat demands, set within the context of continued loss and a lack of available resource could lead to psychological breakdown (3e).

Parents unable to access or accept support, or utilise other secondary coping strategies, may circulate between states of exhaustion and the utilisation of initial coping strategies

(Figure 2.4). The longer-term journey of Dravet necessitates the development of new strategies, however without the space, time, or resources to develop these, parents could become stuck in the cycle of continuous firefighting (3f).

Secondary Coping Strategies

Some parents were in a position to be able to develop secondary coping strategies. This may have been through the availability of resources (3g), or through sheer necessity (3h).

One prominent category was 'receiving and accepting support'. A necessary prerequisite is having support available or offered, which is not the case for many parents. However, when support was available, parents described adjusting between what they may have thought previously (2w), and what was practical, given limited resources, and previous experiences of 'burning out' (3i). There was a reluctant necessity to accepting support as a secondary coping strategy (3j), but an increasing acknowledgement that it was indeed a necessity (3k).

Parents also reported reclaiming some space for themselves, however small this may be, in recognition that this was essential to their mental health, which in-turn affected the care they provide (3h). This interlinks with 'receiving and accepting support', however represents an additional step beyond creating some time by accepting support, in that this time is then utilised to pursue interests (3l), or undertake activities that support mental health (3m). Extended periods of time away from the caring role, despite being uncomfortable and challenging, also yielded significant benefit (3n, 3o).

This time was also used for therapy (3p), the active utilisation of time for oneself, beyond it just being a brief respite, was important (3q), and helped people to reclaim their identity, beyond that of being a carer (3r, 3s).

Integration

Parents who had the opportunity to develop secondary coping strategies had space to integrate the Dravet experience into their lives. This was facilitated by reclaiming an identity beyond the roles associated with caring for a CwDS.

One of the major categories within this theme was 'acceptance'. This was experienced in different ways by parents, with some experiencing it as the cessation or reduction of resentment (3t). Others reported an acceptance of the chronic unpredictability of DS (3u), moving parents to live in the moment (3v).

Distance away from Dravet supported parents' appreciation of their child (3w), which was more difficult if parents were caught in a cycle of continuous firefighting (3x) (Figure 2.4). This appreciation could then be extended to recognising the positive impacts on themselves (3y), as well as other family members, such as siblings (3z). This enabled parents to reflect on and connect with the growth that has occurred during their Dravet journey, which has taught them what is important in life (4a, 4b, 4c), as well as how strong and resilient they are (4d).

Integration does not, however, represent an 'end point'. It is a perhaps best seen as a default state for some parents, from which they can naturally shift when circumstances change. The overarching theme of "lack of resources", paired with the unpredictability of DS means that people can be drawn into other parts of the model at key points of transition (Figure 2.4). For example, during their child's transition to adult services, parents may return to "demands exceed capacity" or indeed the "trauma" parts of this model after a period of acceptance (4e).

Parents who had navigated their way to 'integration' before are equipped to do this again, using the blueprint in their minds (4f), suggesting that there is a protective quality to 'integration', and possessing the armoury of coping strategies that this entails then helps traverse the journey once again (4g).

Table 2.2

Quote Table for Internal Model of Coping and Adjustment

Ref.	Theme	Focussed Category	Quote
2a	Trauma	Single event trauma	"After my son had his second seizure… he nearly died – um, he stopped breathing and the ambulance got lost on the way to hospital. He had to be resuscitated"
2b	Trauma	Single event trauma	"So even after the second seizure they were still quite dismissive, even though he was on life support after that one… I went home… and had PTSD, I still have PTSD after that one"
2c	Trauma	SUDEP	"I think it is the fear of, you know, SUDEP. Because that actually was, when we got the diagnosis…hoping not that one and then of course it was."
2d	Trauma	Secondary trauma	"…watching (CwDS) having a spinal tap…and an MRI when he woke up during it…all of the horrendous life experiences."
2e	Trauma	Secondary trauma	"Because at the time she was so poorly, they did tell me to prepare for the worst. I said could I go to the bathroom please? And I thought I would just dip into the bathroom and I went in the bathroom and just screamed but no noise came out, I just did it silently"
2f	Trauma	Secondary trauma	"I was just in the corner rocking like a maniac… I was like 'oh my god, is he going to die tomorrow?' Awful."
2g	Trauma	Reliving trauma	A: "And I don't think it ever goes" B: "It still gives me goosebumps even" (description of being given diagnosis) B: "Oh, that has given me goosebumps"
2h	Trauma	Dissociation	"Like an out of body experience I think"
2i	Trauma	Reliving trauma	"And even now, but I can't visit it – because I am too traumatised to from it."
2j	Trauma	Recognising trauma	"I always felt like I was having PTSD…going back to the first year of her life. I can't go back there."
2k	Trauma	Recognising trauma	A: "You've gone through a massive trauma" B: "It is trauma, isn't it?" C: "Yeah" D: "It is trauma yeah" E: "Yeah"
2l	Trauma	Repeated trauma	"And the norm was to be in recuss with eighteen people drilling holes into the legs… and that's my line is when the drill comes out and I can't, I can't be in the room"
2m	Trauma	Repeated trauma	"I don't know what I was doing – I was literally living in a state of panic. Walking around with a hospital bag packed the entire time, because you never know...ambulances called to every bloody baby group. People were traumatised."
2n	Trauma	Survival mode	A: "The first six months were like that" B: "And it is like a constant stream of adrenaline actually."
2o	Trauma	Survival mode	"…you go into auto-pilot, do what you need to do – and you're fine, you're fine, you're fine – and then you hit a brick wall a couple of days later and you think, and all your adrenaline rush just drops…I think that is when you start reflecting on what you've been through"
2p	Trauma	Survival mode	"But I was in survival mode…we just needed to get things to survive and help (CwDS)"
2q	Initial coping strategies	Hypervigilance	A: "…when I reflect back, at how much pressure" B: "It was almost obsessive wasn't it?" A: "Aye, it was… couldn't even be alone in the buggy, I'd take her into the kitchen whether it was a cup of tea"

2r	Initial coping strategies	Hypervigilance	A: "Like in the early days…you can't ever relax. You never relax. Even getting in bed at night, you are always prepared to be ready and out. You need to be ready. You have everything ready in the room, monitored up. You're always on stand-by even when you are not." B: "Even when you're asleep. You are not asleep…" A: "You've got one ear and one eye open…"
2s	Initial coping strategies	Becoming a medical professional parent	"I remember reading a study, it was quite a complicated document…but you had to read through all the gumph to think hold on, it could be something"
2t	Initial coping strategies	Becoming a medical professional parent	A: "When she first started having seizures, (partner) was reading, reading like literally everything and…" B: "Obsessed"
2u	Initial coping strategies	Becoming a medical professional parent	A: "I feel like it just took over my life a bit, I was too invested… it became a bit unhealthy…" B: "I did too" A: "It was all I spoke about, it was all I read. Erm, I went through a bad time of it"
2v	Initial coping strategies	Avoidance	A: "I had got to the stage where I wouldn't stop work until ten or eleven at night, and I would leave (partner) sorting it out…but it was because when I was at home it became real, whereas when I was at work, and I could justify being out – I am at work
2w	Unintended consequences	Becoming the only person who can provide care	"I always felt that I knew best and that was it. You know that people didn't have any idea what they were doing with her."
2x	Initial coping strategies	Hypervigilance	"I think time, you spend so much time with him going through all this, it makes you more confident. Definitely. Yeah."
2y	Unintended consequences	Feeling guilty	"…and then she had her very first bad…I had gone away and so I felt guilty, because I wasn't there."
2z	Unintended consequences	Feeling guilty	"I just feel like nothing is working for him, so I feel like I am constantly letting him down."
3a	Initial coping strategies	Dravet before own health	"I lost a lot of weight, just because you don't look after yourself at all. You certainly become number two."
3b	Demands exceed capacity	Having nothing left to give	"I have really struggled a lot, and I think it is probably burnout…"
3c	Demands exceed capacity	Hopelessness	"I went into a really dark place for a few years… it was depression and I was on medication"
3d	Demands exceed capacity	Having nothing left to give	"I went downstairs, bawled my eyes out, composed myself and then went back upstairs and picked her out of the cot, because you're a mum, but you're not superhuman – and sometimes we can't cope."
3e	Demands exceed capacity	Having nothing left to give	"I did have a breakdown about four years ago… I think that was years, years, in the making."
3f	Demands exceed capacity	Having nothing left to give	"And I remember someone saying to me this is a marathon not a sprint…there are bouts of depression from burning out really."
3g	Insufficient resource	Available resource	"We've been very fortunate that…because the actual medical side is pretty much taken care for us…we do get to enjoy her as a child"
3h	Secondary coping strategies	Reclaiming space for self	"And I was like if you don't sort your shit out, you are going to lose your kids, and that was…literally a turning point…I made myself go on a wellbeing course – and it probably was actually was the best thing for me."

3i	Secondary coping strategies	Receiving and accepting support	"So, what has changed, for me, is in the initial days you think nobody can do it as well as you, but as you get older you think well, I can do it better than anybody else, however if I don't get help with this, she'll have to be taken into care because I couldn't manage on my own."
3j	Secondary coping strategies	Receiving and accepting support	"I know I am the best carer for them…but after all those years, I've come to realise that yes I am, but I can't be unless I remove myself for a bit and then go back to it."
3k	Secondary coping strategies	Receiving and accepting support	A: "Yeah, you couldn't manage" B: "So I think that is the biggest change – the journey that you go through, you have got to let people in, and just sort of say, I need help with this. Because, if I don't…" A: "You do actually need help, I was exactly the same."
3l	Secondary coping strategies	Reclaiming space for self	A: "…my sister just signed me back up to a choir four years ago…because I used to love to sing, and I didn't want to go because it was a seven-minute train journey from where (CwDS) was…" B: "You need to… it keeps on giving" A: "I go every week, and it is brilliant. It is a great outlet; singing is very therapeutic. Acceptance, I guess."
3m	Secondary coping strategies	Reclaiming space for self	"When I am running, my head is clear – it'll clean my head, because you need to have that down time to try and control that worry."
3n	Secondary coping strategies	Reclaiming space for self	"You need that time don't you. I mean, you go on a Sunday or a Saturday on your motorbike…I just have my nails done…a bit of self-care…and not feeling guilty about it."
3o	Secondary coping strategies	Reclaiming space for self	"We don't take (CwDS) on holiday with us, it's too stressful for her, and I think that we need a bit of family time away, so she goes into respite."
3p	Secondary coping strategies	Reclaiming space for self	"You might get some therapy…some strategies to deal with it. Even though Dravet is huge, it has to take a back seat for you to deal with the other things that life throws at you."
3q	Secondary coping strategies	Reclaiming space for self	"Now I try really hard all the time every day, I say – what are you going to do for you today, just you."
3r	Secondary coping strategies	Reclaiming identity	A: "I think that protecting your mind and trying not to fall into that…. Doing something that makes you actually feel like you are a person in your own right, and not losing that…" B: "Even that is like self-help though isn't it?"
3s	Secondary coping strategies	Reclaiming identity	A: "It is so important to keep a bit back for yourself, so you are not just your child's mother, father, advocate, professional, nurse, bum-wiper – you still have got…" B: "You've got that identity" A: "You still have got your own."
3t	Integration	Acceptance	And you kind of feel a bit resentful of that too, you have to get through that part of it where you think why has this happened to us? And come to terms with that. It does happen, you do come to terms with it.
3u	Integration	Acceptance	"The worry and the anticipation…that she is going to go into a seizure any minute, that has now gone"
3v	Integration	Acceptance	"You just take each day as it comes, and you are dealt with what you are dealt with…the more you worry, and then worry becomes another worry, and then… you just are forever going back. You just think, I have got to deal with it, it is there – deal with it. Worry about that happens when it happens."
3w	Integration	Finding positives	Interviewer: "What has helped you to live with continued uncertainty?" A: "Celebrating the magic moments"
3x	Demands exceed capacity	Hopelessness	"You have mentioned some really positive things about your child, I can't name one positive thing about mine"
3y	Integration	Finding positives	"My life has changed for the better because of what (CwDS) has brought to my life. She has chilled me out as a person."

3z	Integration	Finding positives	"But I realised as time has gone on again, it has made them into nicer kids…they have got an experience that can't be bought, and I think it is for the better…. I think there is a lot of positives to be taken from it, they have had that experience and they can transfer that into their adult life. They are going to take long-term life lessons from (CwDS) being how she is. In the early days, I didn't see it like that…"
4a	Integration	Growing through the experience	"I have been put in a situation where actually the things that do really matter to you are put at risk, I think that teaches you – or it makes you feel things that you can't feel any other way – so it will teach you lessons that those other things really aren't it."
4b	Integration	Growing through the experience	"It changes you as a person, and the things that were important to you before are not important. We were younger, and we wanted to go on a nice holiday, and we wanted to do this, and they don't matter to you – they are trivial things you think, the things that matter to you now are completely different…"
4c	Integration	Growing through the experience	"And what I do know is, stuff doesn't matter, and I could win the lottery tomorrow and I still cannot fix (CwDS)."
4d	Integration	Growing through the experience	"You are much stronger than what you ever think you are. You are so much more capable of going through the things that you could not possibly manage before receiving the diagnosis…I wouldn't for one minute have believed that I was capable of going through that."
4e	Integration	Acceptance	"I think you put it in a compartment, and there is no room to explore that any more, because I am not going to find the answers that I want and it is not going to change the situation that I am in. You learn to live with it, and learn to deal with it, and learn to find the positive in the situation that we've been dealt with. I mean that's probably where, having an older child. I mean going through to the adult services is a complete rollercoaster…"
4f	Integration	Growing through the experience	"I think it takes a big time for acceptance to come, because I think like I was a totally different person for about four years…now I am a completely different person…a better person…a far more positive person through experiencing these seizures and the things you have seen and lived through…I think it can go two ways; you can either go into yourself and just remove yourself from anything and everything or go right, this is life, how can we make it as normal as possible and I want to start enjoying life again."
4g	Integration	Concrete knowledge	"You have got to pick your battles…if you worry about every little thing, your life comes to a standstill."

4. Discussion

4.1 Language

We were struck by how the phrase 'Dravet journey' was used throughout the focus groups. Parents further along their Dravet journey recognised the challenges of those newer to the journey, whilst often having a foundation of knowledge, skills, and attitudes developed through their own journey that allowed them to see things differently. The present study aimed to capture some of the essence of this process of coping and adjustment.

4.2 Overarching Contextual Themes

The overarching theme of 'lack of resource' reinforces previous research (Jensen et al., 2017) that suggests that parents of CwDS do not have access to sufficient resource, and that this creates a constant struggle. Within this context of a constant struggle, coping and adjustment will be markedly more difficult, and it is likely that families with fewer resources will suffer the greatest impact. Without additional resources provided to these families, many will not have the chance to develop secondary coping strategies.

The overarching theme of 'loss' highlights the continued and repeated loss experienced by parents of CwDS, which is felt from the moment their 'healthy baby died' (1i). Parents are then exposed to repeated stimulus that may reawaken an intense sense of loss, which can be exacerbated by social loss (isolation).

4.3 Model of Coping and Adjustment

The internal model of coping and adjustment provides a theoretical framework for how parents adapt to the relentless demands of caring for a CwDS. The first stage of the model is a period of trauma, which is followed by initial coping strategies and the unintended consequences of these. A cycle is then created between these initial coping strategies and periods of burnout and exhaustion (demands exceed capacity).

Some parents have the opportunity to develop secondary coping strategies, which enable them to integrate the Dravet journey into their lives. These skills allowed parents the space and time to reflect on their personal growth during the Dravet journey, and identify positive aspects of it. These parents will still experience continued loss and encounter insufficient resource.

The final stage of the model (integration) does not represent an end point, as circumstances can draw parents into periods of time when demands exceed capacity, or indeed trauma. We would cautiously predict, however, that parents who have developed secondary coping skills are well equipped to return a state of integration.

4.4 Implications for Practice

The findings from the present study provide an initial steer for the therapeutic interventions that may serve the Dravet community. The identification of pervasive specific trauma phenomena, along with the suggestion that this was not commonly named as trauma within the community is important. This suggests differing levels of trauma recognition, which could have implications as to whether support has been accessed. Future work should be undertaken to raise awareness, develop trauma-specific interventions for parents of CwDS, and ensure professionals in the field work in a trauma informed manner.

The model for coping and adjustment provides a framework for understanding the Dravet journey, and how to support parental integration of the Dravet journey into their lives. Where possible, professionals may be able to work sensitively with parents on the initial coping strategies (e.g. hypervigilance) and the unintended consequences of these to promote long-term parental health. Professionals could also work with parents to develop secondary coping skills, and therapeutically challenge barriers to these (e.g. accepting support). The model may also have a broader practical application beyond Dravet syndrome, such as with parental adjustment to other long-term diagnoses for their children. Additionally, the model could apply to individual experiences of traumatic events that require a degree of coping and adjustment, such as single-event trauma or personally receiving a healthcare diagnosis.

Specific therapeutic work to address loss among parents of children with DS may provide great benefit, considering there is a pervasive recurrent experience of loss and guilt among this population.

4.5 Limitations and Future Research

The present study has a number of limitations. Due to the nature of the sampling (convenience sample), there is inherent bias in the participant pool which may limit the generalisability of the model of coping and adjustment presented.

The sample was also predominantly female (91.7%). Whilst this is consistent with the notion that mothers take on the largest care burden, previous research has suggested that mothers and fathers can bring different approaches to coping with caring for children with significant illness (Ware & Raval, 2007), and so capturing both perspectives is critical. Given the small sample of male participants in the present study, further research should be carried out to explore paternal coping and adjustment. The low male participant numbers may be reflective of an avoidant or supressing coping style (Burden et al., 2016), which, if studied, could yield a different model of coping and adjustment.

The homogenous professional experience (clinical psychology) of the primary researcher and coding pairs presents a contextual consideration, and possible limitation, for the present study. This was in part ameliorated through the methodology, with Charmaz (2006, p 54) stating "careful coding … helps you to refrain from imputing your motives, fears, or unresolved personal issues to your respondents and to your collected data."

4.6 Conclusions

A grounded theory approach was used in the present study to propose a theoretical model as to how parents cope and adjust to caring for a CwDS. We found that there was a significant role of the external factors of loss and lack of resource, which suggest that parents need more support from health and social care. The model of coping and adjustment that we propose had five stages, which are not necessarily met by all parents; trauma, initial coping and unintended consequences, demands exceed capacity, secondary coping strategies, and integration. We hope that this study can be used by

professionals to support families with the integration of the Dravet experience into their lives.

4.7 Declaration of Interests

The authors declare that they have no known competing financial or personal interests.

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Appendix A

(PRISMA Checklist for Systematic Review)

Section and Topic	Item #	Checklist item	Location where item is reported
TITLE			
Title	1	Identify the report as a systematic review.	Title Page
ABSTRACT			
Abstract	2	See the PRISMA 2020 for Abstracts checklist.	Abstract
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of existing knowledge.	1.3 – 1.5
Objectives	4	Provide an explicit statement of the objective(s) or question(s) the review addresses.	1.7
METHODS			
Eligibility criteria	5	Specify the inclusion and exclusion criteria for the review and how studies were grouped for the syntheses.	2.1
Information sources	6	Specify all databases, registers, websites, organisations, reference lists and other sources searched or consulted to identify studies. Specify the date when each source was last searched or consulted.	2.1
Search strategy	7	Present the full search strategies for all databases, registers and websites, including any filters and limits used.	2.1
Selection process	8	Specify the methods used to decide whether a study met the inclusion criteria of the review, including how many reviewers screened each record and each report retrieved, whether they worked independently, and if applicable, details of automation tools used in the process.	2.1; 3.1
Data collection process	9	Specify the methods used to collect data from reports, including how many reviewers collected data from each report, whether they worked independently, any processes for obtaining or confirming data from study investigators, and if applicable, details of automation tools used in the process.	2:1
Data items	10a	List and define all outcomes for which data were sought. Specify whether all results that were compatible with each outcome domain in each study were sought (e.g. for all measures, time points, analyses), and if not, the methods used to decide which results to collect.	Table 1.3
	10b	List and define all other variables for which data were sought (e.g. participant and intervention characteristics, funding sources). Describe any assumptions made about any missing or unclear information.	Table 1.3
Study risk of bias	11	Specify the methods used to assess risk of bias in the included studies, including details of the tool(s) used, how many reviewers assessed each study and whether they worked independently, and if applicable, details of automation tools used in the process.	3.2

Section and Topic	Item #	Checklist item	Location where item is reported
assessment			
Effect measures	12	Specify for each outcome the effect measure(s) (e.g. risk ratio, mean difference) used in the synthesis or presentation of results.	Table 1.3
Synthesis methods	13a	Describe the processes used to decide which studies were eligible for each synthesis (e.g. tabulating the study intervention characteristics and comparing against the planned groups for each synthesis (item #5)).	Figure 1.1; 3.2
	13b	Describe any methods required to prepare the data for presentation or synthesis, such as handling of missing summary statistics, or data conversions.	N/A
	13c	Describe any methods used to tabulate or visually display results of individual studies and syntheses.	N/A
	13d	Describe any methods used to synthesize results and provide a rationale for the choice(s). If meta-analysis was performed, describe the model(s), method(s) to identify the presence and extent of statistical heterogeneity, and software package(s) used.	4.5
	13e	Describe any methods used to explore possible causes of heterogeneity among study results (e.g. subgroup analysis, meta-regression).	N/A
	13f	Describe any sensitivity analyses conducted to assess robustness of the synthesized results.	N/A
Reporting bias assessment	14	Describe any methods used to assess risk of bias due to missing results in a synthesis (arising from reporting biases).	N/A
Certainty assessment	15	Describe any methods used to assess certainty (or confidence) in the body of evidence for an outcome.	N/A
RESULTS			
Study selection	16a	Describe the results of the search and selection process, from the number of records identified in the search to the number of studies included in the review, ideally using a flow diagram.	Figure 1.1
	16b	Cite studies that might appear to meet the inclusion criteria, but which were excluded, and explain why they were excluded.	Figure 1.1
Study characteristics	17	Cite each included study and present its characteristics.	Table 1.2
Risk of bias in studies	18	Present assessments of risk of bias for each included study.	Table 1.2
Results of individual studies	19	For all outcomes, present, for each study: (a) summary statistics for each group (where appropriate) and (b) an effect estimate and its precision (e.g. confidence/credible interval), ideally using structured tables or plots.	Table 1.3
Results of syntheses	20a	For each synthesis, briefly summarise the characteristics and risk of bias among contributing studies.	3.3 – 3.7
	20b	Present results of all statistical syntheses conducted. If meta-analysis was done, present for each the summary estimate and its precision (e.g. confidence/credible interval) and measures of statistical heterogeneity. If comparing groups, describe the direction of the effect.	Table 1.3

Section and Topic	Item #	Checklist item	Location where item is reported
	20c	Present results of all investigations of possible causes of heterogeneity among study results.	N/A
	20d	Present results of all sensitivity analyses conducted to assess the robustness of the synthesized results.	N/A
Reporting biases	21	Present assessments of risk of bias due to missing results (arising from reporting biases) for each synthesis assessed.	N/A
Certainty of evidence	22	Present assessments of certainty (or confidence) in the body of evidence for each outcome assessed.	N/A
DISCUSSION			
Discussion	23a	Provide a general interpretation of the results in the context of other evidence.	4.1; 4.2; 4.3; 4.6
	23b	Discuss any limitations of the evidence included in the review.	4.5
	23c	Discuss any limitations of the review processes used.	4.5
	23d	Discuss implications of the results for practice, policy, and future research.	4.4
OTHER INFORMATION			
Registration and protocol	24a	Provide registration information for the review, including register name and registration number, or state that the review was not registered.	4.8
	24b	Indicate where the review protocol can be accessed, or state that a protocol was not prepared.	4.8
	24c	Describe and explain any amendments to information provided at registration or in the protocol.	Protocol
Support	25	Describe sources of financial or non-financial support for the review, and the role of the funders or sponsors in the review.	N/A
Competing interests	26	Declare any competing interests of review authors.	4.7
Availability of data, code and other materials	27	Report which of the following are publicly available and where they can be found: template data collection forms; data extracted from included studies; data used for all analyses; analytic code; any other materials used in the review.	N/A

Appendix B

(Bennett et al., 2021: CCAT Rating)

	Preliminary	Introduction	Design	Sampling	Data Collection	Ethical Matters	Results	Discussion	Total Score	Precentage	Rating
Bennett 2021	5	5	3	2	4	4	3	5	31	77.5	High

Reference:

Bennett, S. D., Au, C., Byford, S., Chorpita, B., Coughtrey, A. E., Cross, J. H., & Shafran, R. (2021). Feasibility of telephone-delivered therapy for common mental health difficulties embedded in pediatric epilepsy clinics. *Epilepsy & Behavior*, *116*, 107743.

Appendix C

(SRQR Checklist for Major Research Project)

Standards for Reporting Qualitative Research (SRQR)*

http://www.equator-network.org/reporting-guidelines/srqr/

Title and abstract	Location
Title - Concise description of the nature and topic of the study Identifying the study as qualitative or indicating the approach (e.g., ethnography, grounded theory) or data collection methods (e.g., interview, focus group) is recommended	**Title Page**
Abstract - Summary of key elements of the study using the abstract format of the intended publication; typically includes background, purpose, methods, results, and conclusions	**Abstract Page**

Introduction	
Problem formulation - Description and significance of the problem/phenomenon studied; review of relevant theory and empirical work; problem statement	1; 1.1; 1.2
Purpose or research question - Purpose of the study and specific objectives or questions	1.2

Methods	
Qualitative approach and research paradigm - Qualitative approach (e.g., ethnography, grounded theory, case study, phenomenology, narrative research) and guiding theory if appropriate; identifying the research paradigm (e.g., postpositivist, constructivist/ interpretivist) is also recommended; rationale**	2
Researcher characteristics and reflexivity - Researchers' characteristics that may influence the research, including personal attributes, qualifications/experience, relationship with participants, assumptions, and/or presuppositions; potential or actual interaction between researchers' characteristics and the research questions, approach, methods, results, and/or transferability	2.2; 4.5
Context - Setting/site and salient contextual factors; rationale**	2.4
Sampling strategy - How and why research participants, documents, or events were selected; criteria for deciding when no further sampling was necessary (e.g., sampling saturation); rationale**	2.2; 2.5.2
Ethical issues pertaining to human subjects - Documentation of approval by an appropriate ethics review board and participant consent, or explanation for lack thereof; other confidentiality and data security issues	2.1
Data collection methods - Types of data collected; details of data collection procedures including (as appropriate) start and stop dates of data collection and analysis, iterative process, triangulation of	2.4; 2.5.5

sources/methods, and modification of procedures in response to evolving study findings; rationale**	
Data collection instruments and technologies - Description of instruments (e.g., interview guides, questionnaires) and devices (e.g., audio recorders) used for data collection; if/how the instrument(s) changed over the course of the study	2.4; Appendix J
Units of study - Number and relevant characteristics of participants, documents, or events included in the study; level of participation (could be reported in results)	3.1
Data processing - Methods for processing data prior to and during analysis, including transcription, data entry, data management and security, verification of data integrity, data coding, and anonymization/de-identification of excerpts	2.4; 2.5
Data analysis - Process by which inferences, themes, etc., were identified and developed, including the researchers involved in data analysis; usually references a specific paradigm or approach; rationale**	2.5; Figure 2.1; Figure 2.2
Techniques to enhance trustworthiness - Techniques to enhance trustworthiness and credibility of data analysis (e.g., member checking, audit trail, triangulation); rationale**	2.5.5

Results/findings

Synthesis and interpretation - Main findings (e.g., interpretations, inferences, and themes); might include development of a theory or model, or integration with prior research or theory	3.2; Figure 2.4
Links to empirical data - Evidence (e.g., quotes, field notes, text excerpts, photographs) to substantiate analytic findings	Table 2.1; Table 2.2

Discussion

Integration with prior work, implications, transferability, and contribution(s) to the field - Short summary of main findings; explanation of how findings and conclusions connect to, support, elaborate on, or challenge conclusions of earlier scholarship; discussion of scope of application/generalizability; identification of unique contribution(s) to scholarship in a discipline or field	4.1; 4.1; 4.3; 4.4; 4.6
Limitations - Trustworthiness and limitations of findings	4.5

Other

Conflicts of interest - Potential sources of influence or perceived influence on study conduct and conclusions; how these were managed	4.7
Funding - Sources of funding and other support; role of funders in data collection, interpretation, and reporting	4.7

*The authors created the SRQR by searching the literature to identify guidelines, reporting standards, and critical appraisal criteria for qualitative research; reviewing the reference lists of retrieved sources; and contacting experts to gain feedback. The SRQR aims to improve the transparency of all aspects of qualitative research by providing clear standards for reporting qualitative research.

**The rationale should briefly discuss the justification for choosing that theory, approach, method, or technique rather than other options available, the assumptions and limitations implicit in those choices, and how those choices influence study conclusions and transferability. As appropriate, the rationale for several items might be discussed together.

Reference:

O'Brien BC, Harris IB, Beckman TJ, Reed DA, Cook DA. (2014) **Standards for reporting qualitative research: a synthesis of recommendations.** *Academic Medicine*, Vol. 89, No. 9 / Sept 2014 DOI: 10.1097/ACM.0000000000000388

Appendix D

(Ethical Approval)

Dear Dr Liam Dorris
MVLS College Ethics Committee
Project Title *A qualitative exploration of parental adjustment to caring for a child with Dravet Syndrome*
Project No ***200210216***

The College Ethics Committee has reviewed your application and has agreed that there is no objection on ethical grounds to the proposed study.
We are happy therefore to approve the project, subject to the following conditions.

- Project end date as stipulated in original application.
- The data should be held securely for a period of ten years after the completion of the research project, or for longer if specified by the research funder or sponsor, in accordance with the University's Code of Good Practice in Research: (http://www.gla.ac.uk/media/media_227599_en.pdf)
- The research should be carried out only on the sites, and/or with the groups defined in the application.
- Any proposed changes in the protocol should be submitted for reassessment, except when it is necessary to change the protocol to eliminate hazard to the subjects or where the change involves only the administrative aspects of the project. The Ethics Committee should be informed of any such changes.
- For projects requiring the use of an online questionnaire, the University has an Online Surveys account for research. To request access, see the University's application procedure at https://www.gla.ac.uk/research/strategy/ourpolicies/useofonlinesurveystoolforresearch/.
- You should submit a short end of study report within 3 months of completion.

Yours sincerely

Dr Terry Quinn

Terry Quinn
FESO, MD, FRCP, BSc (hons), MBChB (hons)

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Tel – 0141 201 8519

Appendix E

(Minor Amendment Approval)

MA MVLS Ethics Admin
To: Anthony Mercier (PGR)
Cc: [redacted]
Mon 12/12/2022 13:48

Hi Anthony

Sorry for the delay with this. The committee has approved the requested amendments

Regards
Neil

Neil Allan
MVLS Ethics Committee Adminstrator

School of Infection & Immunity
College of Medical, Veterinary & Life Sciences
Glasgow Biomedical Research Centre
Room 314, Sir Graeme Davies Building
University of Glasgow
120 University Place
Glasgow G12 8TA
The University of Glasgow, charity number SC004401

From: Anthony Mercier (PGR) [redacted]
Sent: 25 November 2022 12:42
To: MVLS Ethics Admin <mvls-ethics-admin@glasgow.ac.uk>
Cc: [redacted]
Subject: Minor Ethics Amendment

Hi team,

I hope this finds you well.

This is a request for a very minor change (more of an update) to the ethical approval.

We have ethical approval for the project to take place in London, Manchester and Glasgow - and would like to add Newcastle to this. There was scope for additional locations in the proposal - but this is just to add Newcastle in specifically now that it has been confirmed.

Please find the updated MRP Proposal and Ethics application attached, with this location (Newcastle) added. Do let me know if you have any questions at all.

I have also added the ethical approval confirmation for reference.

Many thanks,

Anthony

MA MVLS Ethics Admin
To: Anthony Mercier (PGR)
Cc: [redacted]
Tue 18/10/2022 13:16

Hi Anthony

Sorry for the delay. The Chair says this is fine to approve.

Regards
Neil

Neil Allan
MVLS Ethics Committee Administrator

Institute of Infection, Immunity & Inflammation
College of Medical, Veterinary & Life Sciences
Glasgow Biomedical Research Centre
Room 314, Sir Graeme Davies Building
University of Glasgow
120 University Place
Glasgow G12 8TA
The University of Glasgow, charity number SC004401

From: Anthony Mercier (PGR)
Sent: 06 October 2022 17:53
To: MVLS Ethics Admin <mvls-ethics-admin@glasgow.ac.uk>
Cc: [redacted]
Subject: Minor Ethics Amendment: 200210216

Dear MVLS Ethics Admin,

Please find a request for a minor amendment to the ethical approval granted for DClinPsy MRP Project (Application number = 200210216).

If it would be possible to fast-track this review, it would be greatly appreciated - as the first focus group is scheduled for this month.

We would like to add the aggregated collection of three pieces of basic demographic data for the cohort - (age range, gender, and years since child's diagnosis).

The responses would be tallied anonymously at each focus group and original document destroyed - meaning that the data would only be collected and stored at aggregate level for the study as a whole, and not linked to any individual participant. Ranged choice options (e.g. 'aged 25-34') also further reduce any possibility of identifying participants from the data.

This amendment would allow for comparisons with previous work that indicates that people who identify as female are more likely to engage in such research, and also provide insight into the sample as a whole (e.g. what was the most common age band of the participants?), which will enhance the conclusions drawn from the research. No data will be stored or presented at individual level.

Please find attached the following that have been added / amended:

- Demographic data tally (new addition)
- MVLS Ethics application (amendments on pages 5 and 8 to include aggregate demographic data)
- MRP Proposal (amendments on page 10 to include aggregate demographic data)
- DPIA (amendments on pages 6, 7, 8, 10 to include aggregate demographic data)
- Participant information sheet (amendments on page 3 to include aggregate demographic data)

If you have any questions at all, or require any further clarification - please do let me know.

Many thanks,

Anthony

Appendix F

(Participant Information Sheet)

This is hosted online in the following location:

https://osf.io/ed4vu

Appendix G

(Privacy Notice)

This is hosted online in the following location:

https://osf.io/3gu7k

Appendix H

(Demographic Survey (Anonymous))

This is hosted online in the following location:

https://osf.io/yxv72

Appendix I

(Consent Form)

This is hosted online in the following location:

https://osf.io/ack6f

Appendix J

(Question Schedule)

This is hosted online in the following location:

https://osf.io/a7d3z

Appendix K

(MRP Proposal)

This is hosted online