

## FULL-LENGTH ORIGINAL RESEARCH

# Child and parental sleep in young children with epilepsy: A population-based case-control study

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## SUMMARY

**Objective:** To determine the prevalence of parent-reported sleep problems in young children with epilepsy and their parents, and to compare findings with those in a non-epilepsy-related neurodisability (neurodevelopmental/neurological difficulties) group.

**Method:** Parents of young children (1–7 years) with epilepsy ( $n = 48$  [91% ascertainment]) completed the Child Sleep Habits Questionnaire (CSHQ). Parents (mothers and fathers) also completed the Pittsburgh Sleep Quality Index (PSQI) and the Iowa Fatigue Scale (IFS) in relation to their own functioning. The responses of parents of children with epilepsy were compared with parents of developmental-, age-, and gender-matched children with nonepilepsy-related neurodisability ( $n = 48$ ).

**Results:** There was not a significant difference in the proportion of children with epilepsy and the children with neurodisability scoring in the at-risk range on the CSHQ (81% vs. 71% respectively) ( $p = 0.232$ ). 62% of mothers and 44% of fathers of children with epilepsy had ‘poor quality sleep’ on the PSQI; there was not a significant difference between mothers of children with epilepsy and those of children with neurodisability ( $p = 0.526$ ) or IFS ( $p = 0.245$ ) total scores. However, mothers of children with epilepsy had significantly more difficulties on the productivity subscale of the IFS ( $p = 0.004$ ). There were no significant differences between fathers’ scores on either measure. In the epilepsy group, child behavioral problems ( $p = 0.001$ ) were independently associated with child sleep difficulties and maternal mental health problems were associated with parental sleep difficulties ( $p = 0.04$ ) and fatigue ( $p = 0.018$ ).

**Significance:** Young children with epilepsy and their parents have a high rate of sleep difficulties. There is a need to develop effective interventions for this population, taking into consideration of the role of child behavioral problems and parental mental health difficulties.

**KEY WORDS:** Epilepsy, Children, Parents, Sleep, Fatigue.



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Although the epilepsies are defined by the occurrence of unprovoked seizures, other factors contribute to diminished quality of life in the children and their families. Cognitive and behavioral difficulties are well described<sup>1,2</sup> and often have a greater impact on health-related quality of life (HRQoL) than seizures.<sup>3</sup> In addition to these difficulties, many children with epilepsy have difficulties with sleep. Disrupted sleep has the potential for negatively impacting children with epilepsy,<sup>4</sup> but also the quality of life of carers. Despite this, sleep difficulties are often not identified or treated.<sup>5</sup>

There is limited population-based data on the prevalence of sleep difficulties in children with epilepsy and their parents, and the majority of previous studies have

## KEY POINTS

- Four of five young children with epilepsy had parent-reported sleep difficulties on a standardized measure
- Sleep difficulties in children with epilepsy were not greater than in an age- and developmentally matched nonepilepsy group
- Mothers and fathers of children with epilepsy reported a high level of difficulties with sleep and fatigue but were not higher than the comparison group
- Child behavioral problems but not epilepsy factors were significantly independently associated with child sleep difficulties
- There is a need to consider both child and parental sleep difficulties in the management of pediatric epilepsy

been hospital based.<sup>6</sup> In addition, few studies have compared sleep in children or their parents with a control group of children with neurodisability (i.e., children with neurologic and/or neurodevelopmental difficulties). It is known that sleep difficulties are common in children with neurodisabilities not associated with epilepsies.<sup>7</sup> It remains unknown whether the presence of seizures in epilepsy has additional impact over and above the impact of associated neurodisabilities. This is important as currently therapeutic approaches in children with epilepsy largely target seizures; this may be appropriate if there is a unique epilepsy contribution to sleep difficulties. In the absence of such a contribution, identification of sleep difficulties in children with epilepsy is necessary so that they can be directly targeted.

The Sussex Early Epilepsy and Neurobehavior (SEEN) project focuses on the prevalence of neurobehavioral difficulties in young children with epilepsy. The aim of the current study was to identify the prevalence of sleep difficulties in young children with epilepsy and their parents. A secondary aim was to compare sleep difficulties in the epilepsy group with a group of age-, gender-, and developmental-matched controls. A final aim was to consider possible contributory factors to sleep difficulties in the children and their parents including demographic factors, epilepsy-specific factors, and child neurobehavior.

## METHODS

### Recruitment

Recruitment in the SEEN study has been described previously.<sup>8</sup> In summary, all children born between 2008 and 2014 in a defined geographic area in the south of the United Kingdom and who were diagnosed with epilepsy (a history of 2 or more unprovoked seizures more than 24 h apart)

between September 30th, 2014 and February 29th, 2016 were eligible. Children needed to be at least 1 year of age during the study period.

### Recruitment of children with epilepsy

A link pediatrician on the research team identified eligible children with epilepsy. All pediatricians, neurophysiologists, and epilepsy specialist nurses in the study area informed the link pediatrician of current/new diagnoses of epilepsy within the study period. The parents/guardians of eligible children were then sent a letter and/or asked in person to complete an interest form if they wished to find out about the study. One of the study psychologists met all agreeing parents to discuss their child's participation.

### Recruitment of children with non-epilepsy-related neurodisability

Once a child with epilepsy was enrolled in the study, parents of children with similar attributes (age, gender, and estimated developmental level) without epilepsy, attending the same clinics in the study area, were approached by collaborating pediatricians. The children required a neurologic or neurodevelopmental concern that needed ongoing intervention. Developmental level was estimated based on school/preschool placement (special or mainstream), previous psychological/developmental assessment, or clinician judgment.

### Psychological assessment

Children in both groups underwent psychological assessment in their homes between November 1st, 2014 and April 30th, 2016. Global development was assessed using the Griffiths Mental Development Scales (GMDS)<sup>9</sup> or Griffiths Mental Development Scales-Extended Revised (GMDS-ER).<sup>10</sup> Behavioral functioning was assessed using the Strengths and Difficulties (SDQ) questionnaire.<sup>11</sup>

Clinical information on all children was extracted using a standardized proforma including data on current AEDs, seizures, and investigations (MRI, EEG). Two pediatric epileptologists independently classified seizures and epilepsy syndromes proposed by the Task Force of the International League Against Epilepsy (ILAE) in 2010.<sup>12</sup> Conflicts were resolved by a third epileptologist. The designation of primary referral concern of the children with non-epilepsy-related neurodisability was based on data extracted on the standardized proforma. Primary referral concerns were collaboratively classified as "global development," "social – communication only," or "motor only" by a pediatrician and child psychologist on the research team.

Socioeconomic Deprivation was determined by the Index of Multiple Deprivation (IMD) 2015 rankings (Department of Communities and Local Government, *English indices of deprivation*. Retrieved from <http://imd-by-postcode.opendatacommunities.org/> [Accessed December 15th, 2016]). Lower scores are associated with lower deprivation. Home

postcodes were used to establish the IMD 2010 ranking of each family.

### Sleep/fatigue assessment in children and parents

Child sleep was measured using the Children Sleep Habits Questionnaire (CSHQ).<sup>13,14</sup> The CSHQ is a parent-completed validated tool for pediatric sleep problems.<sup>13,14</sup> It has 33 items in 8 domains: bedtime resistance, sleep onset delay, sleep duration, sleep anxiety, night wakings, parasomnias, disordered breathing, and daytime sleepiness. Children with total CSHQ scores of  $\geq 41$  are classified as having severe sleep problems.<sup>13</sup> Parents also completed the Early Symptomatic Syndromes Eliciting Neurodevelopmental Clinical Examinations -Questionnaire (ESSENCE-Q).<sup>15</sup> This questionnaire contains one item focusing on the child's sleep. Respondents are asked "Have you (or anybody else) been concerned for more than a few months regarding child's sleep?" The response options are "Yes," "Maybe/A little," or "No."

Caregiver sleep was measured by the Pittsburgh Sleep Quality Index (PSQI).<sup>16</sup> The PSQI assesses both sleep quality and degree of disturbance using 19 items to generate a total score. It consists of 7 subscales: subjective sleep quality, sleep latency, sleep duration, habitual sleep efficiency, sleep disturbances, use of sleeping medication, and daytime dysfunction. Caregivers with total PSQI scores of  $\geq 5$  are classified as having poor quality sleep.<sup>16</sup>

The Iowa Fatigue Scale (IFS)<sup>17</sup> was used to assess parental fatigue. It is an 11-item scale whose items are summed to provide a total score. The items are organized into 4 subscales: cognitive, fatigue, energy, and productivity. The cut-off of  $>35$  was used to identify those with significant fatigue. This cut-off was based on a prior study in which 75% of adults in a primary care setting received scores of  $\leq 35$ <sup>17</sup> and has been used in a previous study of parents of children with epilepsy.<sup>18</sup>

## ANALYSIS

Descriptive statistics were used to characterize scores on the CSHQ, PSQI, and IFS. Chi-square or independent *t*-tests were used to compare the epilepsy and neurodisability groups on child/parent sociodemographic factors. Results of comparisons between groups for the CSHQ, PSQI, and IFS are reported before and after Bonferroni adjustment for multiple comparisons for each set of comparisons.

### Analysis of CSHQ scores

Independent *t*-tests were used to compare mean scores on the CSHQ total score in both groups. Chi-square tests were used to compare the proportions of children in the at-risk range in the epilepsy group compared with the neurodisability group.

Linear regression analyses were performed to identify the factors associated with CSHQ total score in the epilepsy sample and subsequently the total sample. In the epilepsy sample the factors included as possible predictors were gender (male vs. female), child's age (in years), deprivation,

child development score (based on GMDS/GMDS-II), child behavior (SDQ total score), taking sleep medication (yes/no), and the 3 epilepsy factors based on the results of Principal Component Analysis (PCA). PCA (Varimax rotation with Kaiser normalization) was used to reduce the total number of epilepsy factors for the regression analysis. The epilepsy factors included in the PCA analysis were etiology (Genetic/presumed genetic, Structural/metabolic, or Unknown or undetermined), predominant seizure type, seizure frequency (monthly or more often), status epilepticus (seizures longer than 30 min), polytherapy (monotherapy vs. polytherapy), and age at seizure onset (in years), and the analysis resulted in a 3-factor solution accounting for 65% of the variance (see Appendix S1). These 3 factors were subsequently used in the regression analysis. None of the individual epilepsy factors used in the PCA analysis were found to be significant at a 0.05 level.

In the total sample, the epilepsy factors were not included as possible predictors but epilepsy status (present or absent) was included. In the first instance, all independent variables were tested by linear regression. Multivariable analysis was carried out by backward regression, with all predictors entered into the model to identify factors independently associated with the outcome variables.

### Analysis of IFS and PSQI data

#### *Between-group analysis*

Chi-square tests were used to compare the proportions of mothers/fathers in the at-risk range in the epilepsy group compared with the neurodisability group on the PSQI and IFS. Independent sample *t*-tests were used to compare PSQI and IFS scores of fathers/mothers in the epilepsy group compared with the neurodisability groups.

#### *Within-group analysis*

Mean maternal and paternal scores on the PSQI and IFS in both groups were compared with *t*-tests. Independent samples *t*-test were used to compare all mothers and fathers in both groups, whereas paired-sample tests were used to compare mother-father pairs (i.e., children whose mother and father both responded).

#### *Regression analysis*

Multiple regression applying generalized estimating equation (GEE) modeling was used to identify factors associated with total score on the PSQI and IFS in the epilepsy and the total samples. Generalized estimating equation (GEE) is used when there is a possible unknown correlation between outcomes and can be used with related data that arises from correlated data such as family data.

In the epilepsy sample, the 3 epilepsy factors were used in the regression analysis. Additional child factors included were age at time of assessment (in years), gender, global developmental level (based on Developmental Quotient

score from GMDS/GMDS-ER), behavior problems (total SDQ score) and child sleep difficulties (total CSHQ), and deprivation. Parent factors included were respondent (father/mother), mother's age (in years), father's age (in years), maternal education level (formal education/beyond formal education), paternal education level (formal education/beyond formal education), hours worked by mother and hours worked by father and parental mental health (as determined by total score on the Depression Anxiety and Stress Scales – Short Form [DASS-21]).<sup>19</sup>

In the regression analysis for the total sample, additional factors included were epilepsy status (epilepsy/neurodisability). The 3 epilepsy factors were not included in this regression analysis.

Factors associated at the  $p < 0.200$  level were included in the multivariable modeling.

The alpha level for all analyses was  $p < 0.05$ . All analyses were performed with IBM SPSS version 23.0 (Armonk, NY, U.S.A.).

### Ethics approval

The study was approved by the Westminster Research Ethics Committee and was registered with the collaborating hospital primary care organization: The Sussex Community NHS Trust.

## RESULTS

The characteristics of the children in the SEEN study are shown in Table 1 and parental characteristics in Table 2.

During the study period, 53 children with epilepsy were identified in the study area who met eligibility criteria. The prevalence of epilepsy during the study period was 2.7 per 1,000 (1 in 370 95% CI 285–476). Forty-nine parents returned an interest form and the parents of 48 children subsequently completed the CSHQ. Of these 48 children with epilepsy, 47 mothers and 39 fathers completed the PSQI and IFS. In the non-epilepsy-related neurodisability group, 35 children of 48 had a concern about global development

**Table 1. Characteristics of the children in the SEEN study (n = 96)**

	Epilepsy (n = 48)	Neurodisability (n = 48)
Gender (male)	26 (54%)	26 (54%)
Ethnicity (white)	39 (81%)	47 (98%)
Age at time of psych. assessment (mean/range/SD)	4.67 (1–7.16)(1.57)	4.23 (1–7.16) (1.84)
Educational provision (mainstream/special/home)	31/11/6	25/13/10
On SEN register	27(56%)	28 (58%)
Speech and language provision	30 (63%)	35 (73%)
Occupational therapy provision	25 (52%)	25 (52%)
Physiotherapy provision	26 (54%)	33 (69%)
Psychology provision	7 (15%)	7 (15%)
Psychiatry provision	0 (0%)	2 (4%)
Cerebral palsy	7 (15%)	11 (23%)
EEG	48 (100%)	1 (2%)
MRI	34 (71%)	5 (10%)
Duration of epilepsy in years (mean/range/SD)	3.00/0.28–6.52/1.67	N/A
Age of epilepsy onset in years (mean/range/SD)	1.67/0.04–6.00/1.35	N/A
Seizure frequency		
Monthly or more often	32 (67%)	N/A
Less often	16 (33%)	N/A
Predominant seizure type (generalized/focal)	25/23 (52%/48%)	N/A
Electroclinical syndrome	13 (27%)	N/A
Distinctive constellations	1 (2%)	
Structural/metabolic	13 (27%)	
Unknown causes	13 (27%)	
Unclassifiable	8 (17%)	
Genetic/presumed genetic	17 (35%)	N/A
Structural/metabolic	12 (25%)	
Unknown or undetermined	19 (40%)	
Polytherapy	11 (23%)	NA
Seizures longer than 30 min	7 (15%)	N/A
Required rescue therapy	17 (35%)	N/A
Sleep medication	9 (19%)	4 (8%)
ADHD medication	4 (8%)	2 (4%)
Development score (On GMDS/GMDS-ER)	54.38	58.17
Behavioral difficulties (SDQ) <sup>a</sup>	18.16	19.22

N/A, not applicable; SD, standard deviation; SEN, special educational needs; SDQ, Strengths and Difficulties Questionnaire; GMDS/GMDS-ER, Griffiths Mental Development Scales/Griffiths Mental Development Scales-Extended Revised.  
<sup>a</sup>Epilepsy, n = 38; Neurodisability, n = 32.

noted, 7 had a motor concern without reference to developmental delay noted and 15 had social communication difficulties noted (6 of whom did not have developmental delay mentioned as a referral concern). In this group, 56 parents returned an interest form and 48 agreed to their child's participation and completed the CSHQ. Forty-eight mothers and 42 fathers completed the PSQI and IFS in the neurodisability group.

There were no significant differences between children in either group with respect to age ( $p = 0.139$ ), Developmental Quotient ( $p = 0.626$ ), or SDQ total ( $p = 0.494$ ). Among the parents no significant differences were found in maternal age ( $p = 0.230$ ), paternal age ( $p = 0.822$ ), maternal hours worked ( $p = 0.902$ ), or paternal hours worked ( $p = 0.360$ ) between the 2 groups. In addition, there was no significant difference between maternal educational level ( $p = 0.4770$ ), paternal education level ( $p = 0.463$ ), maternal paid employment status ( $p = 0.765$ ), paternal employment status ( $p = 0.596$ ), maternal diagnosis of epilepsy ( $p = 0.153$ ), paternal diagnosis of epilepsy ( $p = 0.233$ ), or paternal mental health diagnosis ( $p = 0.067$ ). The epilepsy group had significantly higher levels of deprivation ( $p = 0.045$ ) and significantly more mothers of children with neurodisability had been diagnosed with a mental health condition than in the group with epilepsy ( $p = 0.04$ ).

Table 3 shows the scores on the CSHQ, PSQI, and IFS for both groups.

### Children's Sleep Habits Questionnaire (CSHQ) and ESSENCE Q-item

In the epilepsy group, 81% of the children had a CSHQ total score  $\geq 41$ . There was no significant difference between the epilepsy group and the neurodisability group (71% scored  $\geq 41$ ) with respect to the proportion of children who scored above the clinical cut-off ( $p = 0.232$ ;  $\chi^2 = 1.429$ ). In addition, there were no significant differences between the groups on the total CSHQ score ( $p = 0.075$ ;  $d = 0.36$ , 95% CI  $-0.04$  to  $0.77$ ) or on any of the subdomains (see Appendix S2). On the sleep item from the ESSENCE-Q, 20 (42%) of parents of children with

epilepsy endorsed "Yes," 8 (17%) "Maybe/A Little," and 20 (42%) "No" regarding concerns about the child's sleep. In the neurodisability group, 19 (40%) of parents of children with epilepsy endorsed "Yes," 5 (10%) "Maybe/A Little," and 24 (50%) "No."

### Parent measures

#### Between-group analysis

There were no significant differences either in the proportions of mothers ( $\chi^2 = 0.017$ ;  $p = 0.898$ ) in each group or fathers in each group with poor quality sleep ( $\chi^2 = 1.010$ ;  $p = 0.217$ ) on the PSQI. There were also no significant differences in mothers' mean total PSQI score between the epilepsy and neurodisability groups ( $p = 0.526$ ), or on any of the subscales or in fathers' mean PSQI scores ( $p = 0.408$ ), or on any of the subscales (see Appendix S3).

The mean scores on the IFS in both groups for mothers and fathers are in Appendix S4. There was no significant differences in the proportions of mothers ( $\chi^2 = 0.274$ ;  $p = 0.601$ ) or fathers ( $\chi^2 = 0.016$ ;  $p = 0.900$ ) in each group with high fatigue on the IFS. Mothers of children with epilepsy scored significantly higher, indicating greater fatigue on the productivity subscale ( $p = 0.004$ ) of the IFS. This difference remained significant after Bonferroni adjustment for multiple comparisons. There was no significant difference between the maternal groups on the total score ( $p = 0.245$ ), cognitive ( $p = 0.845$ ), fatigue ( $p = 0.715$ ), or energy (0.301) subscales of the IFS. There was no significant difference between the fathers on the total scale ( $p = 0.125$ ), or the cognitive ( $p = 0.268$ ), fatigue ( $p = 0.359$ ), energy ( $p = 0.302$ ), or productivity ( $p = 0.62$ ) subscales.

#### Within-group analysis

**Epilepsy group.** In the epilepsy group, there was no significant difference between mothers and fathers on the total PSQI score ( $p = 0.084$ ) or on any of the subscales (see Appendix S5), with the exception of total sleep efficiency where mothers reported significantly more problems

**Table 2. Characteristics of parents in the SEEN study**

	Epilepsy		Neurodisability	
	Mothers (n = 47)	Fathers (n = 39)	Mothers (n = 48)	Fathers (n = 42)
Age	35.29	37.95	33.71	37.61
Deprivation index	16.81	16.81	13.45	13.45
Education level (formal/beyond formal)	23/24	18/21	20/28	16/26
Mean hours worked	8.58	35.32	8.88	37.94
In paid employment	22	35	21	40
Previous diagnosis of epilepsy	2	2	0	0
Previous mental health diagnosis	8 <sup>a</sup>	0	17 <sup>b</sup>	4 <sup>c</sup>

DASS-21, Depression Anxiety and Stress Scales – Short Form.  
<sup>a</sup>Depression (4) Anxiety (2) Both Depression and Anxiety (2) <sup>b</sup>Depression (4) Anxiety (3) Both Depression and Anxiety (8) Bipolar Disorder (2) <sup>c</sup>Depression (4).

**Table 3. Scores on child and parental measures of sleep in the SEEN study**

Scale	Epilepsy	Neurodisability
CSHQ total score (m/SD)	51.69 (10.25)	47.75(11.29)
CSHQ cut-off	39 (81%)	34 (71%)
CSHQ cut-off excluding children <2 year (EP = 45) (NDY = 41)	36 (80%)	32(78%)
PSQI score father (m/SD)	6.36(3.90)	5.71(3.05)
PSQI cut-off for poor quality father	17 (44%)	23 (55%)
PSQI score mother (m/SD)	7.91 (4.28)	7.35(4.31)
PSQI cut-off for poor quality mother	29 (62%)	29(60%)
IFS total score mother (m/SD)	31.13(6.57)	29.54(6.65)
IFS total score father (m/SD)	28.72(6.08)	26.50(6.73)
IFS risk mothers ( $\geq 35$ cutoff)	14(30%)	12(25%)
IFS risk fathers ( $\geq 35$ cutoff)	5(13%)	5(12%)
IFS risk mothers ( $\geq 30$ cutoff)	30 (64%)	24 (50%)
IFS risk fathers ( $\geq 30$ cutoff)	18 (46%)	15 (36%)

CSHQ, Children's Sleep Habits Questionnaire; EP, epilepsy; NDY, neurodisability; PSQI, Pittsburgh Sleep Quality Index; IFS, Iowa Fatigue Scale; m, mean; SD, standard deviation.

( $p = 0.021$ ). This difference was no longer significant after Bonferroni correction for multiple comparisons. There was a higher proportion of mothers in the abnormal range on the PSQI, but this did not reach statistical significance ( $p = 0.094$ ;  $\chi^2 = 2.8106$ ). The paired-samples analysis also did not find a difference between mothers and fathers, with the exception of total sleep efficiency, but this difference did not remain significant after Bonferroni correction for multiple comparisons (see Appendix S6).

On the IFS there was not a significant difference between mothers and fathers in the epilepsy group ( $p = 0.059$ ;  $\chi^2 = 3.565$ ). Mothers had higher scores than fathers on the total scores but this did not reach statistical significance ( $p = 0.084$ ). Mothers reported significantly more fatigue ( $p = 0.013$ ) than fathers on the energy but not the other subscales (see Appendix S5). This difference did not remain significant, however, after adjustment for multiple comparisons. The paired-samples analyses showed that mothers had significantly greater problems on the energy subscale even after Bonferroni correction (Appendix S6).

**Neurodisability group.** In the neurodisability group, mothers or fathers were not significantly more likely to score in the at-risk range on the PSQI ( $p = 0.5879$ ;  $\chi^2 = 0.2936$ ). Mothers had a significantly higher PSQI score than fathers ( $p = 0.04$ ) as well as sleep efficiency score ( $p = 0.03$ ) but not on any of the other subscales (see Appendix S5), but these differences did not remain after Bonferroni adjustment for multiple comparisons. The paired-samples analysis revealed mothers to have significantly more difficulties on the sleep efficiency score after Bonferroni correction (see Appendix S6).

There was no significant difference between mothers and fathers on the IFS at the ( $p = 0.11$ ;  $\chi^2 = 2.51$ , respectively). Mothers had significantly higher scores than fathers on the IFS total score ( $p = 0.03$ ) and the cognitive ( $p = 0.02$ ) and energy subscales ( $p = 0.009$ ) but not on the other subscales. The differences between mothers and fathers remained

significant for the energy subscale after Bonferroni correction for multiple comparisons. The paired-samples analyses showed that mothers had significantly greater problems on the total score, energy, and cognitive subscale even after Bonferroni correction (Appendix S6).

#### Regression analysis

**CSHQ.** In the epilepsy sample, child age, development score, and total score on SDQ were significantly associated with total CSHQ score on univariable analysis (see Appendix S7). On multivariable analysis, the only factor significantly associated with the total CSHQ score was the total score on SDQ ( $p = 0.001$ ). Higher scores on the SDQ (i.e., a greater degree of behavioral difficulty) were associated with higher scores on the CSHQ after adjusting for other factors. In the total sample the only factor associated with total CSHQ was also the total difficulties score on the SDQ on multivariable analysis ( $p < 0.001$ ).

**PSQI.** On the regression analysis in the epilepsy sample the following factors were associated ( $p < 0.200$ ) with PSQI total score (see Appendix S8) on univariable analysis: child gender, developmental quotient, SDQ total score, CSHQ total score, respondent, DASS-21 mother, DASS-21 father, and fathers' education. The only factor significantly associated with total score on multivariable analysis was mothers' DASS-21 score ( $p = 0.040$ ). Greater mental health problems were associated with greater sleep problems. In the total sample (see Appendix S8) the only factors significantly associated with PSQI total score were mothers' DASS-21 score ( $p = 0.01$ ) and respondent ( $p = 0.032$ ). Mothers reported more difficulties than fathers and greater maternal mental health difficulties were associated with higher PSQI scores.

**IFS.** On the regression analysis for the IFS total score in the epilepsy sample the following factors were associated

( $p < 0.200$ ) with total score (see Appendix S9): child age, child gender, developmental quotient, SDQ total score, CSHQ total score, respondent, DASS-21 mothers, DASS-21 fathers, fathers' education, and hours worked (mothers). The only factors significantly associated with total score on multivariable analysis were mothers' DASS-21 score ( $p = 0.018$ ) and respondent ( $p = 0.031$ ). Greater mental health problems were associated with greater fatigue, and mothers reported more fatigue than fathers after adjusting for the other factors.

In the total sample (see Appendix S9), respondent ( $p = 0.002$ ), SDQ total score ( $p = 0.034$ ), and mothers' DASS-21 score ( $p = 0.005$ ) were significantly associated with IFS total score on multivariable analysis. Mothers reported more difficulties than fathers, and greater maternal mental health difficulties and child behavioral problems were associated with increased fatigue.

## DISCUSSION

This study provides population-based data on the high rate of parent-reported sleep difficulties in young children with epilepsy and the significant difficulties with sleep and fatigue experienced by their parents. The inclusion of a neurodisability group matched for developmental status is novel and allowed for a consideration of whether epilepsy uniquely confers an increased risk for difficulties with sleep in children and their caregivers.

Young children with epilepsy have a high rate of parent-reported sleep difficulties, with over half of parents reporting that they were concerned about their child's sleep and 4 of 5 children scoring in the at-risk range on a standardized parent-report measure. Inadequate sleep results in tiredness, difficulties with focused attention, low threshold to express negative affect, and difficulty modulating impulses and emotions in children.<sup>20</sup> Given this, the high level of reported need in the current study suggests that young children with epilepsy should be routinely screened for sleep difficulties. The findings of the current study did not find a difference between the epilepsy group and the neurodisability control group. There were no significant differences between the groups with respect to global development or behavioral difficulties, highlighting that the groups were well matched with respect to neurobehavioral impairment. This suggests that epilepsy does not confer an increased risk for sleep difficulties over and above that conferred by other neurologic/neurodevelopmental difficulties.

This study suggests that sleep is often compromised in parents of children with epilepsy. In one previous study of 52 mothers of children with intractable epilepsy, 67% scored in the "poor sleep" range similar to the 62% in the current study.<sup>21</sup> Almost half of fathers scored in the "poor quality" sleep range in the current study suggesting that difficulties are also common among fathers. Poor sleep quality is associated with a variety of negative consequences in

adults, including health-related problems, diminished quality of life, and economic costs.<sup>22,23</sup> Thus parental sleep difficulties should be considered in the context of family-centered pediatric epilepsy care. The findings of this study add to our understanding of the impact of childhood epilepsy on parental functioning which includes increased symptoms of depression,<sup>24</sup> anxiety,<sup>25</sup> and parenting stress<sup>26</sup> as well as sleep/fatigue difficulties.

In the current study the presence of child behavior problems was the only factor associated with child sleep difficulties. Previous studies have also noted a significant relationship between child behavioral problems and sleep difficulties in pediatric epilepsy.<sup>27,28</sup> The current study did not find a relationship between sleep difficulties and epilepsy factors, which has been shown in previous studies including seizure frequency,<sup>29</sup> AED use,<sup>18</sup> and generalized seizures.<sup>29</sup> Evidence of a relationship between epilepsy factors and sleep from these studies is mixed, but none have been population-based. There are also limited previous studies focusing on contributors to sleep and fatigue difficulties in parents of children with epilepsy. Larson et al<sup>18</sup> reported significantly increased parental sleep difficulties in parents of children with more severe epilepsy and increased number of lifetime AEDs. The lack of a significant association between epilepsy factors and parental sleep in the current study may reflect our population-based approach, age of the children and factors considered as potential contributors. Maternal mental health difficulties were significantly associated with parental sleep and fatigue difficulties suggesting that maternal emotional well-being needs to be considered when developing support for maternal sleep/fatigue difficulties.

### Implications for practice and directions for future research

The high level of difficulties reported in the current study suggests that both child and parental sleep should be considered as part of routine care in pediatric epilepsy clinics. A survey of 17 pediatric neurology providers in the United States found that only one did not routinely ask about sleep problems in children with epilepsy but none routinely used validated sleep questionnaires and that the employed methods underidentified the children at risk for sleep difficulties.<sup>5</sup> Therefore, there is a need to validate instruments such as the CSHQ against results from polysomnography or actigraphy to see if it is a valid measure in children with epilepsy. Given the possible role of behavioral problems with regard to sleep difficulties in childhood epilepsy, there is a need to screen for these problems. Identification and treatment of the behavioral problems may help alleviate sleep difficulties, although in some cases the sleep difficulties may be contributing to the behavioral difficulties and thus need to be prioritized. Likewise parental mental health difficulties need to be taken into account when considering interventions focused on improving parental sleep. The lack of

significant differences between the epilepsy group and the neurodisability group and failure to find an association between epilepsy factors and sleep difficulties suggests that interventions that work for children with other neurologic/neurodevelopmental difficulties may also work in the epilepsy population. However, this hypothesis needs to be tested in robust intervention studies.

### Limitations

The measure of child sleep used is based on parental report only and parental sleep/fatigue is also based on self-report. Although our study was population based, the sample size is small. The children in the study were aged between one and 7 years and our findings may not be of relevance for younger children or children over 7 years of age.

## CONCLUSION

Sleep should be a core concern when considering the psychosocial impact of epilepsy on young children and their families. There is a need to develop effective methods for screening children and to develop evidence-based support for child and parental difficulties. There is also a need to better understand the relationships between child sleep and child behavior as well as parental sleep and parental mental health in this population.

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## DISCLOSURE OF CONFLICT OF INTEREST

Professor Brian Neville was a Trustee of the George E Neville Foundation. None of the remaining authors have any conflict of interest to declare. We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this report is consistent with those guidelines.

## REFERENCES

1. Reilly C, Atkinson P, Das KB, et al. Neurobehavioral comorbidities in children with active epilepsy: a population-based study. *Pediatrics* 2014;133:e1586–e1593.
2. Berg AT, Langfitt JT, Testa FM, et al. Global cognitive function in children with epilepsy: a community-based study. *Epilepsia* 2008;49:608–614.
3. Reilly C, Atkinson P, Das KB, et al. Factors associated with quality of life in active childhood epilepsy: a population-based study. *Eur J Paediatr Neurol* 2015;19:308–313.
4. Stores G, Wiggs L, Campling G. Sleep disorders and their relationship to psychological disturbance in children with epilepsy. *Child Care Health Dev* 1998;24:5–20.
5. Jain SV, Simakajornboon N, Glauser TA. Provider practices impact adequate diagnosis of sleep disorders in children with epilepsy. *J Child Neurol* 2013;28:589–595.
6. Macêdo PJ, de Oliveira PS, Foldvary-Schaefer N, et al. Insomnia in people with epilepsy: a review of insomnia prevalence, risk factors and associations with epilepsy-related factors. *Epilepsy Res* 2017;135:158–167.
7. Didden R, Korzilius HP, Aperlo BV, et al. Sleep problems and daytime problem behaviours in children with intellectual disability. *J Intellect Disabil Res* 2002;46:537–547.
8. Reilly C, Atkinson P, Memon A, et al. Symptoms of depression, anxiety, and stress in parents of young children with epilepsy: a case controlled population-based study. *Epilepsy Behav* 2018;80:177–183.
9. Huntley M. The Griffiths Mental Developmental Scales manual from birth to two years. Bucks: Association for Research in Infant and Child Development. The Test Agency, Thames. 1996:5–39.
10. Luiz D, Faragher B, Barnard A, et al. *Griffiths mental development scales-extended revised: 2 to 8 years—analysis manual*. Oxford, UK: Hogrefe; 2006.
11. Goodman R. The strengths and difficulties questionnaire: a research note. *J Child Psychol Psychiatry* 1997;38:581–586.
12. Berg AT, Berkovic SF, Brodie MJ, et al. Revised terminology and concepts for organization of seizures and epilepsies: report of the ILAE Commission on Classification and Terminology, 2005–2009. *Epilepsia* 2010;51:676–685.
13. Owens JA, Spirito A, McGuinn M. The Children's Sleep Habits Questionnaire (CSHQ): psychometric properties of a survey instrument for school-aged children. *Sleep* 2000;23:1043–1052.
14. Goodlin-Jones BL, Sitnick SL, Tang K, et al. The Children's Sleep Habits Questionnaire in toddlers and preschool children. *J Dev Behav Pediatr* 2008;29:82–88.
15. Hatakenaka Y, Fernell E, Sakaguchi M, et al. ESSENCE-Q – a first clinical validation study of a new screening questionnaire for young children with suspected neurodevelopmental problems in south Japan. *Neuropsychiatr Dis Treat* 2016;12:1739–1746.
16. Buysse DJ, Reynolds CF, Monk TH, et al. The Pittsburgh Sleep Quality Index: a new instrument for psychiatric practice and research. *Psychiatry Res* 1989;28:193–213.
17. Hartz A, Bentler S, Watson D. Measuring fatigue severity in primary care patients. *J Psychosom Res* 2003;54:515–521.
18. Larson AM, Ryther RC, Jennesson M, et al. Impact of pediatric epilepsy on sleep patterns and behaviors in children and parents. *Epilepsia* 2012;53:1162–1169.
19. Henry JD, Crawford JR. The short-form version of the Depression Anxiety Stress Scales (DASS-21): construct validity and normative data in a large non-clinical sample. *Br J Clin Psychol* 2005;44:227–239.
20. Dahl RE. The impact of inadequate sleep on children's daytime cognitive function. *Semin Pediatr Neurol* 1996;3:44–50.
21. Wood LJ, Sherman EM, Hamiwka LD, et al. Maternal depression: the cost of caring for a child with intractable epilepsy. *Pediatr Neurol* 2008;39:418–422.
22. Roth T, Ancoli-Israel S. Daytime consequences and correlates of insomnia in the United States: results of the 1991 National Sleep Foundation Survey. *Sleep* 1999;22(Suppl. 2):S354–S358.
23. Lamberg L. Promoting adequate sleep finds a place on the public health agenda. *JAMA* 2004;291:2415–2417.
24. Ferro MA, Speechley KN. Depressive symptoms among mothers of children with epilepsy: a review of prevalence, associated factors, and impact on children. *Epilepsia* 2009;50:2344–2354.
25. Jones C, Reilly C. Parental anxiety in childhood epilepsy: a systematic review. *Epilepsia* 2016;57:529–537.
26. Cushner-Weinstein S, Dassoulas K, Salpekar JA, et al. Parenting stress and childhood epilepsy: the impact of depression, learning, and seizure-related factors. *Epilepsy Behav* 2008;13:109–114.
27. Cortesi F, Giannotti F, Ottaviano S. Sleep problems and daytime behavior in childhood idiopathic epilepsy. *Epilepsia* 1999;40:1557–1565.
28. Wirrell E, Blackman M, Barlow K, et al. Sleep disturbances in children with epilepsy compared with their nearest-aged siblings. *Dev Med Child Neurol* 2004;47:754–759.
29. Batista BH, Nunes ML. Evaluation of sleep habits in children with epilepsy. *Epilepsy Behav* 2007;11:60–64.



## SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section at the end of the article.

**Appendix S1** Results of Principal Components Analysis using 6 epilepsy factors.

**Appendix S2** Score on CSHQ in SEEN study.

**Appendix S3** Pittsburgh Sleep Quality Index (PSQI) subscale scores in SEEN study.

**Appendix S4** Scores on the Iowa Fatigue Scale in SEEN study – Mother

**Appendix S5** P-values for comparisons between mothers and fathers on the Pittsburgh Sleep Quality Index and Iowa Fatigue Scale subscales in the SEEN study.

**Appendix S6** (a) Paired-samples PSQI – Epilepsy group.  
(b) Paired-samples PSQI – Epilepsy group.

**Appendix S7** Results of univariable and multivariable analysis of factors associated with CSHQ total score.

**Appendix S8** Factors associated with total PSQI score in SEEN study.

**Appendix S9** Factors associated with scores on Iowa Fatigue Scale.