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Sleep disorders and behavioral disorders in Jamaican children with epilepsy: A case–control study

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ABSTRACT

Children with epilepsy (CWE) are more likely to have sleep and behavioral disorders. With differences in reports, the aim was to evaluate sleep and behavior in Jamaican CWE and determine any association with epilepsy-related variables. Children with epilepsy were identified along with age and gender-matched controls from the University Hospital of the West Indies and the Bustamante Hospital for Children. Written informed consent was obtained followed by the completion of questionnaires assessing sleep, behavior, and background of both cases and controls. Parents of 61 children (26 cases and 35 controls) participated in the study. There was no difference in sleep scores or frequency of sleep disorders between cases and controls ($p > 0.05$). Children with epilepsy had worse scores than controls in peer problems (3.85 vs 2.32, $p < 0.01$), internal problems (7.42 vs 4.71, $p < 0.01$), and total behavioral problem (16.27 vs 12.09, $p < 0.01$). When compared with controls, CWE had a higher frequency of abnormal peer problems (66% vs 32%, $p < 0.05$) and emotional problems score (42% vs 15%, $p < 0.05$). Children with learning difficulty were three times more likely to have abnormal behavioral score Odds Ratio (OR) = 3.818, $p < 0.05$). Jamaican CWE have similar sleep scores to their healthy peers but have a higher frequency of peer and emotional problems.

1. Introduction

Comorbidities are often associated with epilepsy and have been shown to affect diagnosis, treatment options, and quality of life [1,2]. This includes sleep disorders and behavioral disorders. Continued exploration of the relationship between epilepsy and sleep has shown that children with epilepsy (CWE) are significantly more likely to have sleep disturbances when compared with the relatively healthy population [3–5]. However, reports have been conflicting. A few studies have shown no difference in sleep problems when comparing patients with epilepsy to age and gender-matched healthy controls [6,7]. While on the other hand, there have been reports of pathological sleep scores in CWE of up to twelve times that as seen in controls [8]. Sleep disorder symptoms reported in CWE include parasomnias, sleep onset delay, and excessive daytime sleepiness [3,8].

Children with epilepsy are also known to have a higher risk for developing behavioral disorders than the relative healthy population [9–11]. The prevalence of behavioral disorders reported in CWE has ranged from 23.7% [12] to 66% [13]. One of the most common behavioral disorders reported in CWE is attention-related problems [10,14]. In a study assessing children with active epilepsy, 80% had behavioral and or cognitive impairment. Of this proportion, 33% had a behavioral diagnosis of attention-deficit hyperactive disorder (ADHD) [11].

Within our population, research has been done to assess cognition comorbidities in CWE [15]; no difference was noted in intelligence quotient (IQ) scores between CWE and controls. However, sleep and behavioral disorders have not yet been studied in this same population. Considering the impact these disorders may have, it was important to assess these parameters in Jamaican CWE.

The aim of the study was to evaluate sleep and behavior in Jamaican CWE and to determine if there is an association with epilepsy-related variables with sleep disorders and behavioral disorders.

Abbreviations: AED, antiepileptic drug; CWE, children with epilepsy; DES, disorder of excessive somnolence; DIMS, disorders of initiating and maintaining sleep; DOA, disorders of arousals; SBD, sleep breathing disorders; SH, sleep hyperhidrosis; SWTD, sleep–wake transition disorders; SDQ, Strengths and Difficulties Questionnaire; SDSC, Sleep Disturbance Scale for Children.

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2. Method

2.1. Study design

The study was a questionnaire-based case–control study. A case was defined as a Jamaican child, age 6–12 years, with a diagnosis of active epilepsy (having experienced seizures within the previous year or currently receiving antiepileptic drug (AED) therapy).

2.2. Recruitment

Cases were recruited from the Paediatric Neurology clinic at the University Hospital of the West Indies and the Outpatient Department general clinic at the Bustamante Hospital for Children in Kingston, Jamaica. Participants of the control group were recruited from the Ophthalmology clinic at the Bustamante Hospital for Children. The Ophthalmology clinic was selected in order to identify relatively healthy children without any chronic illnesses within the hospital setting. Participants were recruited between September 2017 and March 2018. The parents of CWE who were 6–12 years old with uncomplicated, active epilepsy were invited to participate in the study. Uncomplicated epilepsy was defined as epilepsy unassociated with neurologic deficit, metabolic disorder, or severe intellectual disability. Children with epilepsy with any other chronic disease were excluded from the study. Healthy children in the control group were selected and invited to participate in the study based on age and gender matching.

2.3. Instruments

2.3.1. Sleep Disturbance Scale for Children (SDSC) questionnaire

The Sleep Disturbance Scale for Children (SDSC) was developed by Bruni et al. [16] to evaluate sleep behavior and disturbance in children and adolescents in the past 6 months. It was initially validated on 1157 children from ages 6 to 15 years with a reported sensitivity of 0.89 and a specificity of 0.74.

The SDSC contains 26 questions rated on a Likert scale ranging from 1 to 5 (1 = never, 2 = occasionally, 3 = sometimes, 4 = often, 5 = always). Severity increases with increased scores. The following sleep disturbance factors can be assessed: sleep-related breathing, initiating and maintaining sleep, arousal, sleep–wake transition, excessive somnolence, and sleep hyperhidrosis (SH). The total raw scores range from 38 to 130. A score sheet is given with raw scores and their corresponding standardized T-score; a T-score of greater of 70 is abnormal. The SDSC has been used in recent studies assessing sleep in CWE and has been able to identify sleep disturbances in this population [8,17].

The SDSC was adjusted and validated for the Jamaican population. The questionnaire was issued to 10 randomly selected parents to complete. Parents were asked to comment on any questions that were difficult to understand and to make recommendations on how to improve these questions. The questionnaire was then adjusted and issued to 10 randomly selected parents to complete. The same adjusted questionnaire was reissued to the same parents after two weeks to ensure internal consistency.

2.3.2. Strengths and Difficulty Questionnaire (SDQ)

The Strengths and Difficulties Questionnaire (SDQ) was used to assess behavior. It is a widely used screening tool for behavioral problems in children ages 4–17 years. It is a 25-item questionnaire that has five scales — emotional symptoms, conduct problems, hyperactivity/inattention, peer relationship problems, and prosocial problems. Questions are scored on a Likert scale (not true, somewhat true, and certainly true). The total difficulties score is determined by the sum of all scales except prosocial problems. This score ranges from 0 to 40. The internalizing score is a sum of emotional and peer relationship scales (ranges from 0 to 20) while the externalizing score is a sum of conduct and hyperactivity scales (ranges from 0 to 20). Scores were previously categorized as normal, borderline, and abnormal where a higher score is indicative of greater severity of symptoms except in the case of the prosocial scale [18].

A more recent four-tier classification has been used where scores are classified as close to average, slightly raised (or slightly lower), high (low), or very high (very low). Low scores refer to the prosocial scale that is rated inversely. The SDQ has been recently used in CWE [12] and has been previously used within the Jamaican population [19].

2.4. Procedure

The researcher, who was not a part of the clinical team, invited parents/guardians of eligible children to participate after being identified by clinical staff. Parents/guardians who chose to participate signed an informed written consent and completed questionnaires (which was done using a structured interview format) of the SDSC and the SDQ. This was done in a quiet area by the researcher. A demographic questionnaire was also completed by the parent/guardian. Completion of questionnaires lasted approximately 10–15 min. Clinic staff extracted information related to the patient's medical history from the dockets of CWE whose parents consented to participate in the study. This included epilepsy classification, AED treatment, and seizure characteristics (onset, frequency, etc.).

2.5. Data analysis

The data were analyzed using Statistical Package for the Social Sciences (SPSS) version 20. All variables were tested for normality in distribution using the Shapiro–Wilks method. A comparison of sleep variables and behavior variables was done between cases and controls. For normally distributed data, continuous variables were analyzed using the independent t-test and categorical variables using Pearson's chi-square test. Otherwise, the Mann–Whitney and Fisher's exact test were used accordingly. Spearman's correlation test was used to determine the association between variables. Regression analyses were performed to determine predictors of sleep and behavior. Multiple binary logistic regression was used for binary categorical dependent variables whereas linear regression was done for continuous dependent variables. Statistical significance was taken at $p < 0.05$.

2.6. Ethical consideration

Ethical approval was granted for the study from the University of the West Indies Faculty of Medical Sciences Ethics Committee (ECP 78,16/17) and the South East Regional Health Authority of the Ministry of Health, Jamaica.

3. Results

3.1. Demographics of participants

Parents of 61 children (26 cases and 35 controls) consented to take part in the study. The mean Standard Deviation (SD) age of CWE was 8.88 (1.77) years; 58% were males. The mean Standard Deviation (SD) age of controls was 8.60 (1.65) years; 54% were males. There was no significant difference between cases and controls with regard to age and sex (Table 1). No child had been previously diagnosed with a sleep disorder. Four (15%) CWE had been previously diagnosed with a behavioral disorder. Three (75%) had ADHD. One (3%) child in the control group had been previously diagnosed with autism. There was no significant difference between CWE and controls in learning difficulty based on parent perception or cosleeping (Table 1).

Table 1
Demographics of cases and controls.

Variables	Cases (n = 26)	Controls (n = 35)	p-Value
Male, n (%)	15 (57.7)	19 (54.3)	0.794
Age (years), $\bar{x}$ (SD)	8.88 (1.77)	8.60 (1.65)	0.584
Previous diagnosis			
Sleep disorder, n (%)	0	0	–
Behavior disorder, n (%)	4 (15.4)	1 (2.9)	0.099
Asthma, n (%)	1 (3.8)	8 (22.9)	0.039
Cosleep, n (%)	20 (76.9)	23 (65.7)	0.343
Parent perception of learning			
Learning difficulty, n (%)	13 (50)	9 (25.7)	0.051

3.2. Clinical features of the group with epilepsy

Twenty-one (80.8%) CWE had been diagnosed with a focal epilepsy. Overall, seizure types present in our CWE were focal with impaired awareness (47.6%), focal to bilateral tonic–clonic (23.8%), generalized onset tonic–clonic (14.2%), focal aware with motor onset (4.8%), generalized motor tonic–clonic (4.8%), and generalized onset myoclonic (4.8%). Electroclinical syndromes were identified in 28.6% of the CWE. Syndromes identified were childhood epilepsy with central temporal spikes (19%), frontal lobe epilepsy (4.8%), and childhood absence epilepsy (4.8%).

Parents were asked if at least 75% of the seizures occurred during the day or at night, during sleep. Nineteen (73%) parents reported that their children had nocturnal seizures. One (3.8%) parent reported that there was no obvious time of day when seizures occurred. Thirteen (50%) had not had seizures within the last six months prior to the parent being interviewed. Seven (26.9%) had one or more seizures within the last six months prior to the interview but less frequently than once per month (Table 2).

Twenty-two (84.6%) cases were being treated with monotherapy at the time of the study. Carbamazepine and valproic acid were the AEDs most frequently used (50% and 33%, respectively). The time within which they had been last prescribed the drug ranged from six months to more than a year.

Table 2
Clinical characteristics of children with epilepsy.

Variable	Mean (SD)	Range
Seizure onset (years)	4.32 (3.19)	0.5–11.0
Seizure duration (years)	4.57 (2.88)	0–11.0
	n (%)	
Seizure type		
Focal	21 (80.8)	
Generalized	5 (19.2)	
Seizure frequency (in last 6 months)		
Daily	1 (3.8)	
Weekly	3 (11.5)	
Monthly	2 (7.7)	
Less than 1 per month	7 (26.9)	
None	13 (50)	
Seizure diurnal pattern		
Night	19 (73.1)	
Day	6 (23.1)	
Both	1 (3.8)	
Drug therapy		
None	2 (7.7)	
Monotherapy	22 (84.6)	
Polytherapy	2 (7.7)	
Current AED		
Monotherapy		
Carbamazepine	12 (50)	
Valproic acid	8 (33.2)	
Lamotrigine	1 (4.2)	
Levetiracetam	1 (4.2)	
Polytherapy		
Carbamazepine and valproic acid	1 (4.2)	
Phenobarbital and levetiracetam	1 (4.2)	

AED, antiepileptic drug.

3.3. Sleep Disturbance Scale for Children (SDSC)

3.3.1. Validity of SDSC

Ten participants were included in the test–retest of the SDSC. One participant (10%) was excluded from analysis as they were not available for retesting. One question was adjusted. Question 25 was changed from "The child experiences daytime somnolence" to "The child experiences daytime sleepiness".

Bland Altman plots were used to determine fixed bias or systematic difference between the mean and difference of the scores of subscales of the SDSC measured before and after adjustment. For all six subscales (disorders of initiating and maintaining sleep (DIMS), sleep breathing disorders (SBD), disorders of arousals (DOA), sleep–wake transition disorders (SWTD), disorders of excessive somnolence (DES), and SH) and Total SDSC score, there was no fixed bias between test and retest measurements ($p > 0.05$).

3.3.2. SDSC scores

Nine participants with asthma were excluded during analysis of the SDSC scores (1 case, 8 controls). The SDSC scores of 52 (85%) participants, 25 cases, and 27 controls were analyzed.

The scores for the subscales of sleep disorders were similar for cases and controls ($p > 0.05$). There was also no significant difference in median total SDSC score ($p = 0.720$) or the prevalence of abnormal scores for cases and controls (Table 3).

The frequency of sleep disorders ranged from 0 to 20%. The sleep disturbance with the highest frequency of abnormal scores was that of DES (Table 3).

In further analysis, CWE were divided into two groups based on their AED therapy. The two most predominantly used drugs, carbamazepine (50%) and valproic acid (33%) were used. No difference was seen between CWE on carbamazepine and CWE on valproic acid in sleep scores.

There was no significant association between epilepsy-related variables and sleep scores.

3.4. Strengths and Difficulties Questionnaire (SDQ)

Data on the SDQ from 60 (98%) participants, 26 cases and 34 controls, were analyzed. Data from one (2%) participant from the control group were excluded as insufficient questions were answered.

3.4.1. Mean SDQ scores

The mean total score for the SDQ was significantly higher in cases when compared with controls ($p = 0.005$). The mean score for peer problems was significantly higher in CWE when compared with controls ($p = 0.002$). The internal problems score, which is a combination of the emotion problems score and the peer problems score, was also significantly higher in CWE ($p = 0.003$) than in controls. No significant difference in conduct or hyperactivity scores was noted between groups (Table 3).

Table 3
Comparison of median and abnormal SDSC and SDQ scores between cases and controls.

	SDSC median scores, (IQR)			Frequency of abnormal SDSC scores, n (%)		
	Cases (n = 25)	Control (n = 27)	p-Value	Cases (n = 25)	Control (n = 27)	p-Value
DIMS	10 (3)	11 (4)	0.360	2 (8)	2 (7.4)	0.665
SBD	4 (3)	3 (2)	0.606	3 (12)	3 (11.1)	0.628
DOA	3 (1)	3 (0)	0.518	0 (0)	1 (3.7)	0.519
SWTD	9 (5)	8 (5)	0.567	3 (12)	2 (7.7)	0.481
DES	9 (5)	7 (6)	0.200	5 (20)	4 (14.8)	0.449
SH	2 (0)	2 (0)	0.758	1 (4)	2 (7.4)	0.529
Total SDSC score	39 (10)	37 (16)	0.720	2 (8)	2 (7.7)	0.680
	SDQ mean scores, $\bar{x}$ (SD)			Frequency abnormal SDQ scores, n (%)		
	Cases (n = 26)	Control (n = 34)	p-Value	Cases (n = 26)	Control (n = 34)	p-Value
Emotion problems	3.58 (2.72)	2.38 (2.12)	0.07	11 (42.3)	5 (14.7)	0.017
Conduct problems	3.08 (1.83)	2.29 (2.05)	0.131	11 (42.3)	10 (29.4)	0.299
Hyperactivity problems	5.77 (2.50)	5.09 (2.76)	0.328	6 (23.1)	8 (23.5)	0.967
Peer problems	3.85 (1.95)	2.32 (1.74)	0.002	17 (65.4)	11 (32.4)	0.011
Prosocial problems	7.85 (2.29)	8.26 (2.62)	0.520	5 (19.2)	4 (11.8)	0.422
Total SDQ score	16.27 (5.34)	12.09 (5.71)	0.005	12 (46.2)	8 (23.5)	0.065
Internal problems	7.42 (3.70)	4.71 (3.19)	0.003			
External problems	8.85 (3.44)	7.38 (4.19)	0.153			

DIMS, Disorders of initiating and maintaining sleep; SBD, sleep breathing disorders; DOA, disorders of arousal; SWTD, sleep–wake transition disorders; DES, disorders of excessive somnolence; SH, sleep hyperhidrosis; IQR, Interquartile Range; Internal problems score is a sum of emotion and peer problems; External problems score is a sum of conduct and hyperactivity problem.

There was correlation between age ($r(60) = 0.272$, $p < 0.05$), epilepsy status ($r(60) = 0.345$, $p < 0.01$), learning difficulty based on parent perception ($r(60) = 0.451$, $p < 0.001$), and total SDSC score ($r(51) = 0.568$, $p < 0.001$) with mean total SDQ score. Further assessment of the internal problem using Spearman's correlation analysis showed that age ($r(60) = 0.349$, $p < 0.01$), learning difficulty based on parent report ($r(60) = 0.382$, $p < 0.01$), and epilepsy status ($r(60) = 0.364$, $p < 0.01$) correlated with this subscale of the SDQ scores. Multivariate regression analysis identified age ($B = 0.631$, $p = 0.014$), learning difficulty based on parent report ($B = 1.874$, $p = 0.038$), and epilepsy status ($B = 2.145$, $p = 0.013$) as significant independent predictors for higher scores in the internal problems subscale.

3.4.2. Abnormal SDQ scores

Participants who scored high or very high on the SDQ subscales (or low or very low in the case of prosocial problems) were classified as having an abnormal score.

When compared with controls, CWE had a significantly higher frequency of abnormal emotion problem scores (42% vs 15%, $p = 0.017$) and abnormal peer problems (65% vs 32%, $p = 0.011$) (Table 3).

The relationship between demographic variables (age, sex), epilepsy and asthma status, learning according to parent report, previous behavioral disorder diagnosis and cosleep, and abnormal SDSC scores and abnormal total SDQ score was examined using Spearman's correlation. Age ($r(60) = 0.310$, $p = 0.016$) and learning according to parent report ($r(60) = 0.342$, $p = 0.007$) were weakly correlated with abnormal SDQ score. Multivariate logistic regression indicated that children with learning difficulty are almost three times more likely to have abnormal behavioral scores ($\beta = 3.818$, $p < 0.05$) (Table 4).

There was no significant correlation of epilepsy status with abnormal scores for peer, emotion, or total SDQ scales.

Table 4
Predictors of having abnormal score on the SDQ of cases (n = 26) and control (n = 34).

Variable	Odds ratio (β)	95% CI	p-Value
Age	1.469	1.000–2.157	0.050
Learning difficulty[a]	3.818	1.166–12.505	0.027

CI, Confidence Interval.
[a] Based on parent perception.

4. Discussion

In this study, we found that Jamaican children with uncomplicated epilepsy had similar sleep scores on the SDSC questionnaire when compared with their peers. Our results differ from the study reported by Gutter et al. [8], which involved 130 children with partial epilepsy and 166 matching controls. In this study, all parameters of the SDSC questionnaire were significantly higher in the CWE. Similarly, Samaitiene et al. [20] examined sleep disorders in 20 children with rolandic epilepsy and found significantly a higher total sleep disorder score, attributable to higher SWTD and DES when compared with the healthy controls. Interestingly, Samaitiene et al. identified loss in these differences when the study included children without active seizures in the past three months. This disparity was also reported by Cortesi et al. [21] who investigated sleep in a group of 89 children with both partial and generalized seizures and 321 healthy controls. They found that sleep problems occurred significantly more in CWE. Again, this difference was lost the longer these CWE were seizure-free. In our study, parents of more than half of the CWE reported no seizures within the last six months prior to the study onset. This may account for the absence of sleep disorders in CWE in our study.

The frequency of abnormal sleep scores in CWE reported in this study is considerably lower than reported frequencies in other studies. In both developed and developing countries, frequencies of abnormal sleep scores have ranged from 29% to 80% [8,17,22]. We also found no association between epilepsy-related variables, such as epilepsy type, seizure frequency, AED therapy and diurnal patterns, and sleep problems. This has similarly been reported in other studies [23]. The low frequency of abnormal sleep scores in this study corroborates with the previous diagnosis of sleep disorders by parent report. No participant within the study had been previously diagnosed with a sleep disorder. It may be possible that parents of CWE are aware of the role sleep plays in epilepsy and are already taking precautions to maintain good sleep hygiene.

While there were no significant differences in the frequencies of abnormal sleep disorder scores, it was noted that the sleep disorder with the highest frequency of abnormal scores was DES. Disorders of excessive somnolence is one of the most often reported sleep disorders in CWE. In previous studies, DES was associated with nocturnal seizures [24]. However, in this study, no difference was seen

between the different diurnal patterns (nocturnal vs daytime seizures) in CWE.

We assessed risk factors, such as cosleeping and AED therapy, which have been reported in other studies [4,25]. We found that cosleeping was not significantly different between cases and controls and did not prove to be a risk factor for sleep disorders in CWE. Antiepileptic drugs did not prove to be a risk factor for sleep disturbances. However, the number of CWE not on AEDs in our study was too small for comparison with children on AED therapy.

The SDQ was used in the study to assess behavior. Jamaican CWE in our study had higher total scores on the SDQ. In addition, almost half of Jamaican CWE in this study had abnormal behavioral scores compared with approximately 25% of controls. Our findings are at the higher end of the range reported by a recent International League Against Epilepsy (ILAE) Task Force Report. By examining epidemiological studies done worldwide, they concluded that typically 35 to 50% of CWE have behavioral problems [26].

Children with epilepsy had significantly worse scores compared with controls in the subscales of peer problems and the internal problems score, which combines the subscales of peer problems and emotional problems. There was also a significant difference between cases and controls in the frequency of abnormal scores in emotional problems and peer problems. Studies have reported both emotional and peer problems present in CWE [12,27]. However, they are always reported to occur alongside other behavioral problems of a more external nature such as conduct problems, hyperactivity problems, or attention problems.

To our knowledge, no study has reported on the presence of only internalizing problems in CWE. In our study, age was found to be an independent predictor of the internal problems score. Older age has been shown to be associated with internalizing problems in CWE [28]. In a health status report, Fox et al. [29], suggested that there was a cause for concern about the emotional well-being in Jamaican teenagers (aged 10 to 15 years) as 14% had behaviors considered as internal problems (suicidal thoughts, feeling withdrawn). There were no teenagers involved in our study; however, 34% were 10–12 years old. It may be that Jamaican CWE exhibit behaviors typically associated with older children [27]. The reason for this is uncertain, and a more in-depth analysis should be done to determine the reason for this higher likelihood of internal behaviors in Jamaican CWE.

While no formal assessment of cognition was done, learning difficulty was assessed by parent report. The difference in learning difficulties between CWE and controls did not meet statistical significance; however, there was a notable difference. Furthermore, learning difficulties based on parent report was an independent predictor of higher SDQ scores and abnormal SDQ score in the general sample. Our findings show that children with learning difficulties are 3.8 times more likely to have behavioral problems. This is similar to reports in literature where cognitive impairment was shown to be associated with behavioral problems [30,31]. No significant independent predictors were identified in the group with epilepsy possibly because of the small sample size.

4.1. Strengths and limitations

This is the first study to assess sleep disorders and behavioral disorders in Jamaican CWE. The use of age and gender-matched controls allowed us to control for these confounding factors. The results of the study, however, cannot be generalized as the sample size was small.

4.2. Conclusion

In this study, Jamaican CWE were found to have similar sleep scores when compared with controls. However, more CWE had behavioral problems, particularly internal type — peer and emotional problems. These findings suggest that Jamaican CWE should be screened for behavioral problems, particularly internalized behaviors.

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Declaration of competing interest

None.

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