

SLEEP-RELATED PROBLEMS IN CHILDREN WITH CEREBRAL PALSY AND ITS CORRELATION WITH SLEEP DISTURBANCE IN PRIMARY CAREGIVERS

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Abstract

Background: Sleep-related issues can present additional challenges for children with cerebral palsy (CP). This study investigated sleep problems in children with CP and their correlation with primary caregivers attending a tertiary care centre. **Materials and Methods:** This is a cross-sectional observational study conducted at Paediatric OPD, Neurology OPD and DEIC OPD of Govt. RMH, Thanjavur, Tamil Nadu, India from Jan 2019 to Dec 2019. Children aged 6-15 years with CP and their primary caregivers were included. A validated sleep questionnaire was used to assess sleep-related problems. Data were analyzed using descriptive statistics and chi-square tests. **Results:** The study included 70 children with CP (55.72% male) and their caregivers. Most children had spastic quadriplegia (40%) and a history of seizures (74.28%). Sleep disturbances were prevalent among both children and caregivers. Disorders of initiating and maintaining sleep (DIMS) had a prevalence of 31.43%, followed by sleep breathing disorders (SBD) at 21.43%. Sleep-wake transition disorders (SWTD) were observed in 10% of the participants. Significant associations were found between sleep disturbance in children and their caregivers, particularly for DIMS, SBD, and sleep hyperhidrosis (SHY) scores. **Conclusion:** Sleep disturbances are common in children with cerebral palsy and have an impact on their caregivers. This study highlights the interconnectedness of sleep disruption between children with CP and their caregivers. Understanding and addressing sleep-related issues in this population can improve the overall well-being and quality of life for children and their caregivers. Further research and interventions are needed to manage sleep problems in children with cerebral palsy effectively.

INTRODUCTION

Sleep is an essential aspect of human life, constituting approximately one-third of our time. It is a natural, cyclical state in which our responsiveness decreases, and we emerge from it spontaneously. In the case of children and adolescents, good quality sleep plays a crucial role in their physical and mental growth and development. Sleep-related issues can pose additional challenges for individuals with cerebral palsy, a group of permanent disorders that affect movement and posture development.^[1] Cerebral palsy (CP) is characterized by damage to the developing brain's motor pathways, resulting in difficulties with movement and posture. It is a non-hereditary and non-progressive condition but exhibits a dynamic clinical pattern despite a static pathology. While cerebral palsy is not curable, it can be improved through training and interventions. Therapies and rehabilitation aim to enhance

functional abilities, maximize potential, and improve the quality of life for individuals with cerebral palsy.^[2,3] CP manifests from early childhood or infancy. Cerebral palsy is characterized by persistent motor impairments without regression. A family history of similar neurodegenerative diseases can indicate a specific genetic condition, setting it apart from cerebral palsy, which is not typically associated with specific genetic causes. Movement disorders such as ataxia, characterized by uncoordinated movements, are commonly observed in neurodegenerative diseases but are not typically associated with cerebral palsy. Lastly, abnormal body odours and other distinctive features, such as a cherry red spot, can be specific to certain neurodegenerative disorders, further aiding in their differentiation from cerebral palsy.^[4] Its prevalence varies worldwide, with population-based studies reporting rates ranging from 1.5 to > 4 cases per 1,000 live births or children within a specific age range. On a global scale, the overall

birth prevalence of cerebral palsy is estimated to be around 2 per 1,000 live births. However, this prevalence can differ across regions. For example, the prevalence ratio falls from 1.3 to 4.4 cases per 1,000 live births in countries like India, China, and Turkey. Cerebral palsy is commonly observed in preterm infants and is strongly associated with periventricular leukomalacia.^[5-7] This condition involves damage to the immature oligodendroglia between 20 and 34 weeks of gestation. The damage is often caused by oxidative stress resulting from ischemic (hypoxic-ischemic encephalopathy) or infectious or inflammatory insults.

In light of these challenges, a study was conducted in Tamil Nadu, a state in India, aiming to investigate sleep-related problems in children with cerebral palsy. The study also explored the correlation between sleep disturbances in these children and their primary caregivers attending a tertiary care center.

MATERIALS AND METHODS

This cross-sectional observational study was conducted at the Department of Paediatrics, Govt. RMH, Thanjavur Medical College, Thanjavur, a tertiary care teaching hospital in South India, from January 2019 - December 2019. Seventy children aged 6-15 years were selected randomly from the CP children attending Paediatric OPD, Neurology OPD and DEIC OPD of Govt. RMH, Thanjavur. A validated sleep questionnaire measured sleep-related problems in CP children and their caregivers.

Inclusion Criteria

Children and adolescents between the age of 6 and 15 years, both genders, diagnosed with cerebral palsy, primary caregiver present with the patient and able to provide detailed antenatal and perinatal history, and CP children who are on a stable dosage of antiepileptic, antispastic or any other drug which can cause sedation in the last six months were included.

Exclusion Criteria

Co-morbid health problems like a cardiorespiratory disease or any other illness that could affect health, CP children/caregivers diagnosed with depression or other psychiatric or chronic medical conditions, etc., which may affect sleep patterns, the inability of the parent to provide medical information, change in the medication within the past six months that could

affect sleep pattern, unreliable history, and hypotonic/floppy child were excluded.

The parents/caregivers of the CP children who fulfilled the inclusion criteria and consented to participate in the study were interviewed. A detailed history of the CP child, including antenatal and perinatal history, developmental history, history of other systemic illness, H/O seizure and drug history, was obtained from the parents/caregivers. The motor disability of the child was classified using GMFCS into five levels. The data required for SDSC and PSQI questionnaires were obtained by interviewing the parents/caregivers by the investigating paediatrician of the study.

Statistical Analysis

Data were entered in Microsoft Excel and analyzed using the IBM Statistical Package for Social Sciences (SPSS) software version 19.0. Continuous data such as age, SDSC and PSQI Quantitative data were presented in numbers and percentages in tables and bar charts. Heterogeneity chi-square or Fisher's exact test was applied to find the association between Sleep disturbance in children and their caregivers and other variables such as type of CP, GMFCS levels etc. Pearson's correlation was performed to find the strength of the association between sleep disturbance in children and their caregivers. It was represented with the 'r' value. A p-value < 0.05 was considered statistically significant.

RESULTS

In the present study, a majority (55.72%) of the participants were boys and male to female ratio was 1.25:1. The mean age of the study participants was 9.107±2.25. The minimum age was 6.5 years, and the maximum was 13 years. Most children were between 6.5 and 10 years (64.29%); 35.71% were between 10 and 13 years. The maximum number of patients belongs to GMFCS level 4(40%). There were no patients with GMFCS level 1. The majority of the children had Spastic quadriplegia (40%), followed by Spastic diplegia (28.57%) and spastic hemiplegia (25.72%). A history of seizure was present in 74.28% of the children. 25.72% of the children did not have a history of seizures. Most children (74.29%) have a history of bed-sharing with their caregivers. 25.71% of the children did not have a history of bed sharing with their caregivers (Table 1).

Table 1: Demographic and sleep characteristics of children with cerebral palsy

		Number	Percentage
Gender	Male	39	55.72
	Female	31	44.28
Age category	<10 years	45	64.29
	>10 years	25	35.71
GMFCS level	Level 2	7	10
	Level 3	15	21.43
	Level 4	28	40
	Level 5	20	28.57
Type of Cerebral Palsy	Spastic quadriplegia	28	40

	Spastic diplegia	20	28.57
	Spastic hemiplegia	18	25.72
	Extrapyramidal	4	5.71
H/O Seizure	Present	52	74.28
	Absent	18	25.72
Bed Sharing	Present	52	74.29
	Absent	18	25.71
Hours of sleeping	9 - 11 hrs	31	44.26
	8 - 9 hrs	12	17.14
	7 - 8 hrs	18	25.71
	5 - 7 hrs	9	12.86
	< 5 hrs	0	0
Minutes taken to fall asleep	< 15	24	34.29
	15 - 30	16	22.86
	30 - 45	5	7.14
	45 - 60	5	7.14
	> 60	20	28.57

Table 2: Frequency of sleeping problems reported in caregivers using the PSQI questionnaire

Components	Component Scores			
	0	1	2	3
Subjective sleep quality	8	46	7	9
Sleep latency	28	12	10	20
Sleep duration	24	24	11	7
Habitual sleep efficiency	46	16	0	8
Sleep disturbances	10	55	5	0
Use of sleeping medication	69	1	0	0
Daytime dysfunction	50	5	11	4

Table 3: SD in children, according to SDSC

	SD in children, according to SDSC							SD in caregivers, according to PSQI
	DIMS	SBD	SWTD	DA	DES	SHY	TS	
Score range	7-27	3-10	6-19	3-7	5-17	2-10	27-78	1-16
Mean	13.528	4.128	8.928	3.657	7.885	3.671	41.8	5.585
Standard deviation	6.394	1.872	2.965	0.976	3.491	2.775	13.282	4.65
Cut off	>16	>6	>13	>5	>12	>6	>51	>5
Normal (n, %)	48 (68.57%)	55 (78.57%)	63 (90%)	64 (91.42%)	60 (85.71%)	62 (88.57%)	53 (75.72%)	47 (67.14%)
Pathological (n, %)	22 (31.43%)	15 (21.43%)	7 (10%)	6 (8.58%)	10 (14.29%)	8 (11.43%)	17 (24.28%)	23 (32.86%)

The prevalence of disorders of initiating and maintaining sleep (DIMS) among the study participants was 31.43%, with a mean score of 13.528 ± 6.394 (7-27). Sleep breathing disorders (SBD) prevalence was 21.43% with a mean score of 4.128 ± 1.872 (3-10). The prevalence of sleep-wake transition disorders (SWTD) was 10%, with a mean score of 8.928 ± 2.965 (6-19). The prevalence of disorders of arousal (DA) was 8.58%, with a mean score of 3.657 ± 0.976 (3-7). The prevalence of disorders of excessive sleepiness (DES) was 14.29%, with a mean score of 7.885 ± 3.491 (5-17). The prevalence of Sleep Hyperhydrosis (SHY) was 11.43%, with a mean score of 3.671 ± 2.775 (2-10) (Table 3).

Table 4: Associations between sleep disturbance severity scores (SDSC) and various sleep parameters in children with cerebral palsy

	PSQI Score	Normal sleep (%)	Disturbed sleep (%)	Total	P value
SDSC	Normal Score	44(83.01%)	9(16.99%)	53(100)	<0.001
	Pathological Score	3(17.64%)	14(82.36%)	17(100)	
SDSC - DIMS	Normal Score	41(85.42%)	7(14.58%)	48(100)	<0.001
	Pathological Score	6(27.28%)	16(72.72%)	22(100)	
SDSC - SBD	Normal Score	39(70.90%)	16(29.10%)	55(100)	0.198
	Pathological Score	8(53.33%)	7(46.67%)	15(100)	
SDSC - DIMS	Normal Score	41(85.42%)	7(14.58%)	48(100)	<0.001
	Pathological Score	6(27.28%)	16(72.72%)	22(100)	
SDSC - SWTD	Normal Score	45(71.42%)	18(28.58%)	63(100)	0.02
	Pathological Score	2(28.58%)	5(71.42%)	7(100)	
SDSC - DA	Normal Score	42(65.62%)	22(34.38%)	64(100)	0.377
	Pathological Score	5(83.33%)	1(16.67%)	6(100)	
SDSC - SHY	Normal Score	46(74.20%)	16(25.80%)	62(100)	0.0004
	Pathological Score	1(12.50%)	7(87.50%)	8(100)	

The study investigated the relationship between sleep disturbance in children with CP and sleep disturbance in their caregivers. Different assessment scores were used to evaluate various aspects of sleep. The findings revealed significant associations between sleep disturbance in children and their caregivers in several domains. Firstly, among CP children with normal total sleep scores, most (83.01%) of caregivers experienced normal sleep, while a minority (16.99%) reported disturbed sleep. Conversely, among CP children with pathological total sleep scores, a significant majority (82.36%) of caregivers experienced disturbed sleep, while only a small proportion (17.64%) had a normal sleep. Similar patterns were observed for the DIMS score, where caregivers of CP children with pathological scores reported significantly higher rates of disturbed sleep (72.72%) than those with normal scores (14.58%).

Moreover, the SWTD score also showed a significant association, with caregivers of CP children with pathological scores experiencing higher rates of sleep disturbance (71.42%) than those with normal scores (28.58%). On the other hand, the DES and DA scores did not demonstrate significant associations with sleep disturbance in caregivers, indicating that these particular aspects of sleep in CP children may not strongly impact their caregivers' sleep patterns. However, the SHY score, which assesses sleep hyperhidrosis, revealed a significant association, with a majority (87.50%) of caregivers of CP children with pathological scores reporting disturbed sleep. These findings emphasize the interconnectedness of sleep disruption between CP children and their caregivers, highlighting the potential impact of CP-related sleep issues on the well-being of the child and their primary caregiver.

Table 5: Associations between sleep disturbance and various factors in children with cerebral palsy

		NS		PS		P value
		N	%	N	%	
Age	<10 years	32	71.12	13	28.88	0.228
	>10 years	21	84	4	16	
Gender	Male	28	71.8	11	28.2	0.39
	Female	25	80.65	6	19.35	
Type of CP	Spastic quadriplegia	16	57.15	12	42.85	0.02
	Spastic diplegia	18	90	2	10	
	Spastic hemiplegia	15	83.33	3	16.67	
	Extrapyramidal	4	100	0	0	
GMFCS Level	Level 2	7	100	0	0	0.448
	Level 3	11	73.33	4	26.67	
	Level 4	21	75	7	25	
	Level 5	14	70	6	30	
History of seizure	Present	36	69.23	16	30.77	0.0315
	Absent	17	94.44	1	5.56	
Bed sharing	Yes	34	65.38	18	34.62	0.594
	No	13	72.22	5	27.78	

The current study examined the relationship between sleep disturbance and various factors in children with cerebral palsy (CP). The findings revealed that age was not significantly associated with sleep disturbance in CP children ($p=0.228$). Similarly, no significant statistical association was found between sleep disturbance and gender in CP children ($p=0.390$). However, the study did identify a noteworthy correlation between sleep disturbance and the specific type of cerebral palsy known as spastic quadriplegia ($p=0.02$). This suggests that children with spastic quadriplegia may be more prone to experiencing sleep disturbances compared to those with other forms of cerebral palsy. Furthermore, a significant association was observed between a history of seizures and sleep disturbance in children with cerebral palsy ($p=0.0315$). This implies that CP children with a history of seizures are likelier to experience sleep disturbances than those without (Table 5).

DISCUSSION

Cerebral palsy is the most common chronic motor disability that begins in early childhood. There is a higher frequency of sleep problems in children with cerebral palsy than those without chronic health conditions. Various studies have shown the prevalence of sleep disorder in school-aged CP children to vary widely from 15-40%.⁸⁻¹⁴ In our study, males outnumbered females as 55.72% of the participants were boys and Male: Female ratio was 1.25: 1. Similar findings were reported in Newman et al. study with male preponderance (57.8%) and Male: Female ratio, 1.36:1.^[8] However there was a female preponderance in Munyumu et al. study conducted at Uganda and Adiga et al. study conducted at Bangalore, India.^[9,10]

Our study included CP children between 6.5 and 15 years, as the Sleep Disturbance Scale for Children was originally validated for children aged 6.5 to 15.3.12. In the study, most children (64.29%) were between 6.5 and 10 years, whereas 35.71% were between 10 and 13 years. The mean age of the study population was 9.1 years, with a standard deviation

of 2.25 years. Similarly, in the study by Newman et al., the mean age of the participants was 8 years 10 months with SD 1 year 11 months.^[8] In the Adiga et al. study, the mean age was 107.9 months (8.9 years) with SD 29.5 months.⁹

The severity of motor disability based on GMFCS was as follows: Level 2- 10%, Level 3- 21.43%, Level 4- 40%, and Level 5- 28.57%. In a similar study by Newman et al., it was reported that the GMFCS level of children was Level 1- 42.2%, Level 2-19.1%, Level 3 – 17.3%, Level 4 - 13.3%, and Level 5- 8.1%.⁸ GMFCS levels among the study population of Adiga et al. study were Level 1 – 40%, Level 2 – 28%, Level 3 -12%, Level 4 -2%, Level 5 - 18%.^[9]

In our study, most children had a history of seizure (74.28%) and were on AEDs. However, we included only the children who did not have seizure episode in the past 6 months and is on a stable dosage of antiepileptic and antispastic drugs. About 25.72% of the children had no history of seizures and were not on antiepileptic treatment. We categorized the total score and scores of individual sleep disorders obtained by the SDSC questionnaire into pathological and normative data for the scale. A T score of more than 70 was considered pathological, and T score of 70 or less was regarded as the normal range. Based on the SDSC assessment, our study showed that about 24.28% of the children (n=17) had total pathological scores. The most common sleep disorder was DIMS which was present in 31.43% of the children in this study. In contrast to the findings of this study, Adiga et al. reported total pathological scores as high as 36% of the children and DIMS in as high as 50% of the CP children in their study.^[9] Newman et al. found the prevalence of SD in their study as 22.5%. About 24.3% of children had DIMS in his study.^[8] In this study, assessment of other subscales of SDSC revealed Sleep breathing disorders in 21.43% of the children, Disorders of excessive sleepiness in 14.29%, Sleep hyperhidrosis in 11.43%, Sleep-Wake transition disorders in 10%, Disorders of arousal in 8.58% of the children. We determined the factors associated with sleep disturbance in CP children using baseline characteristics and biomedical factors such as age and sex of the child, type of CP, the severity of motor disability (GMFCS), and the presence or absence of epilepsy. Among children < 10 years of age, 28.88% and among children > 10 years of age, 16% had a total pathological sleep score. Among the boys, 28.20% and the girls, 19.35%, had total pathological sleep scores. Statistical analysis showed no significant association between age (p=0.228) or gender (p=0.390) with sleep disturbance in CP children. There was no statistically significant association between sleep disturbance and GMFCS level in this study, as an increase in the level of functional impairment as recorded by GMFCS did not significantly increase the sleep disturbance in CP children (p=0.448).

Similarly, in the study done by Adiga et al., no statistically significant association was found between sleep disturbance and GMFCS level in CP children.^[9] However, in contrast to our study, in the study by Domenico M. Romeo et al., a significant association was found between sleep disturbance and level 5 on the GMFCS. Children having epilepsy had a significant association with total pathological sleep score (p=0.0315). Similar findings were found in the studies done by Adiga et al.^[9], Munyumu et al.^[10] and Domenico M. Romeo et al.^[14] Among the types of cerebral palsy, spastic quadriplegia was more associated with sleep disturbance (42.85%). There was a significant association between spastic quadriplegia and total pathological sleep score (p=0.02). The caregivers' Sleep disturbance was assessed using the PSQI questionnaire, which assesses sleep quality and disturbance over a 1-month interval. A PSQI score of > 5 was considered pathological. In our study, 32.86% of the caregivers had pathological PSQI scores. Sleep disturbance in primary caregivers was significantly associated with total sleep score (p<0.001), DIMS (p<0.001), SWTD (p=0.02) and SHY (p=0.0004) in children. Bed sharing was not significantly associated with pathological sleep scores in caregivers (p=0.594).

The correlation between sleep disturbance in children with cerebral palsy and sleep disturbance in their caregivers was positive and statistically significant (r value-0.69). The present study showed the prevalence of sleep-related problems in CP children and its significant association with sleep disturbance in primary caregivers.

Limitations

Being a single-centred study and including only the children who reach the tertiary care centre, findings may not represent the general population. The study did not include children under 6.5 years and > 15 years. Other limitations include not matching the data with age-matched controls, relying on parental reports due to lack of self-reporting vision, not performing IQ tests, etc.

CONCLUSION

Around one-fourth of the CP, children had pathological sleep in the study. About one-third of the mother had sleep disturbance in the study. There was a significant association between SD in children (TS, DIMS, SWTD and SHY) and their respective caregivers. There was a significant positive correlation between CP children's sleep disturbance and their caregivers' sleep disturbance. Among the types of CP, Spastic quadriplegia had a significant association with SD. The presence of epilepsy in CP children had a significant association with SD. The association between SD in CP children and GMFCS levels, age and gender was insignificant. Bed sharing had no significant association with SD in caregivers.

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