Prevalence of Sleep Disorders in Children with Cerebral Palsy; A Questionnaire-based Observational Study

Shilpa Kulkarni, Tanmay Sanjay Jadhav

Department of Paediatric Neurosciences, Bai Jerbai Wadia Hospital for Children, Parel, Mumbai, Maharashtra, India

Background: Cerebral palsy (CP), the most common developmental disorder, has many comorbidities (epilepsy and behavioral issues). Sleep disturbances are common complaints of parents and are usually neglected in pediatric populations in comparison to other well-documented co-morbidities of CP but may have a significant effect on the quality of life of children and their parents. Objective: To study the prevalence and pattern of sleep disorders (SD) in children with CP. Study Design: Questionnaire-based observational study. Setting: Pediatric tertiary care center. The study was done over 6 months. Participants: In total, 200 children with CP between the ages of 1 year to 14 years were included in the study by convenience sampling. Co-morbid health problems involving cardiorespiratory system, other illness (e.g. epilepsy and gastroesophageal reflux disease), or children on anticonvulsant medications altering sleep patterns were excluded. Intervention: The Sleep Disturbance Scale for Children (SDSC) was administered to assess the presence of pathological sleep and type of SD ranging between Disorders of Initiation and Maintenance of Sleep (DIMS), Sleep Breathing disorders (SBD), Disorders of Arousal (DA), Sleep-Wake Transition Disorders (SWTD), Disorders of Excessive Somnolence (DES), and Sleep Hyperhydrosis (SHY). Main Outcome Measures: Sixty-two percentage of children (124) had a pathological total sleep score (score >39). Results: DIMS are the most common, occurring in 78.2% of subjects with a pathological sleep score (score >39). Quadriplegics [n = 96, mean score = 49.86(16.38)] and GMFCS V [n = 19, mean score = 58.00(17.10)] are most severely affected.

Conclusion and Discussion: Children with CP have under-reported SD and DIMS is the most common type of SD. There is a linear correlation between the extent of topographical and motor afflictions and SD.

Keywords: Cerebral palsy, children, questionnaire, sleep, sleep disorders

INTRODUCTION

Cerebral palsy (CP) is defined as a heterogenous group (dynamic disorder) of motor impairment syndromes caused by a nonprogressive (static) lesion of the developing brain. In India, its prevalence is around three cases per 1000 live births; however, being a developing country, the actual figure may be much higher than the probable figures.[1] CP, predominantly a motor disorder, also has many comorbidities like epilepsy, gastrointestinal (GI) disturbances, and behavioral problems. From clinical experience, it has been noted that, in addition to motor impairments and related disabilities, there is an increased frequency of various sleep disorders (SD). Very few research have been conducted in India on these lines. SD definitely require...
a systematic study as its appropriate management may have a beneficial effect on the quality of life of children with CP and their parents. As literature regarding the same is scarce from Indian subpopulation, we were determined to conduct this study.

**Aims**
To study prevalence and correlate the types of SDs with various classifications of cerebral palsy.

**SUBJECTS AND METHODS**

**Subjects**
This retrospective observational study was conducted at The Bai Jerbai Wadia Hospital for Children between January and May 2018 after approval from the Institute’s ethics committee.

Two hundred children clinically diagnosed as CP, visiting developmental outpatient clinics or physiotherapy departments, and who are in the age groups of 1 year to 14 years and primary caregivers who consented to participate in the study were included. Unreliable history, comorbid health problems involving cardiorespiratory system, or any other illness like epilepsy and GERD, which may alter sleep patterns, were excluded. The patients who were on sleep-altering medications like anticonvulsants were also excluded. Demographic and clinical details were noted for all the patients in the study. Their neurological and functional status were noted. Children with CP were on any medications (anti-spasticity, anti-dystonia, antiepileptics) were noted. Children with CP were classified according to neurological, topographical, and functional classification at the time of evaluation.

**Clinical evaluation**
GMFCS scores were used to quantify the subjects’ motor functional state. This includes five functional levels (Level I to Level V, in the decreasing order of functional capabilities) with a description of skills for five different age groups including <2 years, 2–4 years, 4–6 years, 6–12 years, 12–18 years. Generic neurological (spastic, dystonic, and mixed) and topographical (monoparesis, diparesis, hemiparesis, paraparesis [residual], and quadriparesis) classifications were used to further categorize the subjects.

Sleep Disturbance Scale for Children (SDSC) questionnaire was administered to assess SD in study subjects. Permission for using the SDSC questionnaire was sought from the author of the original article validating the questionnaire. The SDSC assesses sleep quality and disturbance in the children in the last 6 months. It contains 27 items and covers 6 general and common sleeping disorders in children: Disorders of initiation and Maintenance of sleep (DIM), Sleep Breathing disorders (SBD), Disorders of Arousal (DA), Sleep-Wake Transition Disorders (SWTD), Disorders of Excessive Somnolence (DES), Sleep Hyperhydrosis (SHY). The total sleep score as well as individual disorder score were calculated. Total score and scores of individual SD were categorized into pathological and normal based on normative data from the scale used[1] (SDSC).

A total score of more than 39 was considered pathological as this has been proven to be the diagnostic cut-off from the scale. These scores were correlated with different types of CP (topographical and neurological).

**Statistical analysis**
Variables from the SDSC were correlated with parameters of demographic and GMFCS score. Collected data were tabulated and analyzed using the Statistical Package for Social Science (SPSS; IBM). Various tests such as the NPar test, Kruskal–Wallis, and Spearman’s test were used for understanding correlations and associations between factors and variables.

**Results**
Of the total of 200 subjects, the mean age was 3.6(2.5) years. There were 139 males [mean total score = 46.31(13.77)] and 61 females [mean total score = 48.59(15.35)]. The mean total score of all subjects was 47.05(14.27). The difference in total scores is not statistically significant between genders (P = 0.395), indicating that the sleep problem is not affected significantly by the sex of the individual.

The subjects as per neurological classification of CP were categorized into spastic, dystonic, and mixed types, and the total scores were compared. The mean total score, among Spastic group, was 45.20(11.76) [n = 83; 41.5%] and dystonic group was 50.90 (17.910 [n = 42; 21%] and mixed was 54.81(10.33) [n = 75; 37.5%]. The difference was statistically significant between groups by ANOVA test (P = 0.008), indicating that the type of affliction had an impact on the extent of sleep disturbance with mixed having the highest mean score.

By topographical classification, the sample had three with monoparesis [mean total score = 30.25(1.35)], 20 with Hemiparesis [mean total score = 41.85(7.95)], 71 with diparesis [mean total score = 43.38(11.99)], 10 with (residual) paraparesis (at the time of assessment) [mean total score = 47.5(12.45)], and 96 with quadriparesis [mean total score= 49.86(16.38)].
The total sleep score between groups is statistically significant \((P = 0.019)\), indicating that the sleep is affected greater as the area of affection is increased with a correlation coefficient of 0.19.

Total analysis of the mean scores of children with respective to topographical classification is given in Table 1.

Twenty-two children were GMFCS level I \([\text{mean total score} = 47.68(13.47)]\), 76 GMFCS level II \([\text{mean total score} = 43.55(11.86)]\), 52 GMFCS level III \([\text{mean total score} = 43.86(11.19)]\), 31 GMFCS level IV \([\text{mean total score} = 53.83(17.65)]\), and 19 GMFCS level V \([\text{mean total score} = 58.00(17.10)]\).

The mean total scores had a significant statistical association with GMFCS scores \((P < 0.05)\) but with a weak correlation \((\text{coefficient} = 0.267)\), indicating increased sleep disturbance with increasing disability.

The SD were classified into six subcategories according to the scale used \(\text{(in patients with pathological scores):} \) Disorders of Initiation and Maintenance of Sleep \(\text{(DIMS)}, \) Sleep Breathing Disorders \(\text{(SBD)}, \) Disorders of Arousal \(\text{(DA)}, \) Sleep-Wake Transition Disorders \(\text{(SWTD)}, \) Disorders of Excessive Somnolence \(\text{(DES)}, \) and Sleep Hyperhydrosis \(\text{(SHY)}\).

The percentage of children with individual category of SD \(\text{(out of 124 pathological sleep scores)}\) is given in Table 2.

Disorders of initiation and maintenance of sleep is most commonly present with 78.2\% \((n=97)\) of all children with a pathological sleep scores \((n=124)\) affected by it.

The total score is positively correlated with each subtype of sleep disorder, indicating that an increasing total score leads to increased scores of each subtype and hence an increased affliction \([\text{Table 3}]\).

The strongest correlation exists between T-score and DIMS \((\text{correlation coefficient: 0.635)}\). Weak correlations exist between T-score and DES \((\text{coefficient: 0.465)}, \) SWTD \((\text{Ccoefficient: 0.409)}, \) SBD \((\text{coefficient: 0.336)}, \) and DA \((\text{coefficient: 0.234).}\)

**DISCUSSION**

The prevalence of sleep problems is estimated to be 20\% to 42\% across childhood\(^1\) in non-CP children according to recent research. Children with CP are prone to sleep problems with an incidence recently reported to be 23\%,\(^2\,3\) The difficulty may include one or a combination of many types of sleep problems like problems of initiating and maintaining sleep, sleep breathing problems, excessive sleep sweating, etc.

| Total score | N | Mean | Std. deviation | Std. error |
|-------------|---|------|----------------|------------|
| Monoparesis | 3 | 30.250 | 1.3500 | 0.9462 |
| Hemiparesis | 20 | 41.850 | 7.9556 | 1.7789 |
| Diparesis   | 71 | 45.380 | 11.9969 | 1.4238 |
| Paraparesis | 10 | 47.500 | 12.4566 | 3.9391 |
| Quadriparesis | 96 | 49.865 | 16.3852 | 1.6723 |
| Total       | 200 | 47.055 | 14.2752 | 1.0094 |

**Table 2: Percentage of CP children affected by different types of sleeping disorders**

| Severe (\% ) | DIMS, % | SBD, % | DA, % | SWTD, % | DES, % | SHY, % |
|--------------|---------|--------|-------|---------|--------|--------|
| Mild         | 13.7    | 2.4    | 9.7   | 9.7     | 7.3    | 2.4    |
| Moderate     | 36.3    | 10.5   | 19.4  | 29.8    | 16.9   | 5.6    |
| Severe       | 28.2    | 20.2   | 15.3  | 4.8     | 5.6    | 24.2   |
| Conjugate total | 78.2   | 33.1   | 44.4  | 44.4    | 29.8   | 32.3   |

**Table 3: Correlation coefficient of the total score against different types of sleeping disorders**

| DIMS | SBD | DA | SWTD | DES | SH |
|------|-----|----|------|-----|----|
| 0.635| 0.336| 0.234| 0.409| 0.465| 0.191|

Key 1: Types of sleeping disorders: DIMS = Disorders of Initiation and Maintenance of Sleep; DA = Sleep Breathing Disorders \(\text{(SBD)}, \) Disorders of Arousal; SWTD = Sleep-Wake Transition Disorders; DES = Disorders of Excessive Somnolence; SHY = Sleep Hyperhydrosis
Sleep problems in CP children are under-reported and undertreated in comparison to clinical experience.[4] Our aim was to assess the extent of sleep problems in children with CP using a validated sleep questionnaire for children (SDSC).[5] Among the multiple questionnaires available for assessing sleep quality,[6] this was the right choice for our current study on the basis of it being able to assess various aspects of sleep without having the child's pathological condition as a variable. The validity of the questionnaire has been studied by multiple researchers.[7-9] 62% of the children were affected with varying degrees of sleep problems. Children suffered most from DIMS (78.2% of children with a pathological sleep score) and least from excessive somnolence (29.8% among affected). Newman et al.[2] found DIMS to be the most common subtype of sleep disorder among spastic quadriplegics. We studied the correlation between degree of sleep disturbance and extent of topographical involvement of the CP and found a positive correlation. Hence, quadriplegics were most severely affected. We also found a positive correlation between the sleep disturbance with GMFCS scoring. Hence, GMFCS V were most severely affected. Newman et al.[2] reported that the principal factor associated with total sleep disturbance was the presence of active epilepsy, yet the exact association of epilepsy and SD is not well understood yet. Gender was not statistically significant to the total sleep score. Many recent studies have been conducted to associate sleep and quality of life (QOL) between CP children and their normal peers. Insomnia, DES, and decreased functional ability were associated with lower QOL in children with CP. This indicates the importance of assessing sleep quality and disorders in patients with CP and adequately responding to their needs clinically.[10,11] The limitation of the study was the fact that it was only questionnaire-based and polysomnography for the subjects was not done. It would be ideal to follow up this study with one that is based on polysomnography of children who are identified to have a pathological sleep score.

In summary, the results of our study indicate that CP children have a higher incidence of SD than previously reported and hence missed clinically. The most common form of SD across age, topographical, and GMFCS classification is DIMS most likely due to the inability of acquiring a comfortable sleep position and physical pain due to muscle spasms or abnormal muscle movement.[12] There is a positive correlation of degree of sleep disorder with increasing severity of CP in terms of the extent of topographical affection (especially quadriplegics) and GMFCS grading (especially at grade IV and V). The SDSC can be used routinely as a simple screening tool for CP patients with suspected sleep problems.

Financial support and sponsorship
Nil.

Conflicts of interest
There are no conflicts of interest.

References
1. Bharti B, Mehta A, Malhi P. Sleep problems in children: A guide for primary care physicians. Indian J Pediatr 2013;80: 492-8.
2. Newman CJ, O'Regan M, Hensey O. Sleep disorders in children with cerebral palsy. Dev Med Child Neurol 2006;48:564-8.
3. Elsayed RM, Hasanean BM, Sayyah HE, El-Auoty MM, Tharwat N, Belal TM. Sleep assessment of children with cerebral palsy: Using validated sleep questionnaire. Ann Indian Acad Neurol 2013;16:62-5.
4. Dominic AF, Jennifer F, Peter PV. Assessing and managing lung disease and sleep disordered breathing in children with cerebral palsy. Paediatr Respir Rev 2009;10:18-24.
5. Bruni O, Ottaviano S, Guidetti V, Romoli M, Innocenzi M, Cortesi F, et al. The sleep disturbance scale for children (SDSC). Construction and validation of an instrument to evaluate sleep disturbances in childhood and adolescence. J Sleep Res 1996;5:251-61.
6. Spruyt K, Gozal D. Pediatric sleep questionnaires as diagnostic or epidemiological tools: A review of currently available instruments. Sleep Med Rev 2011;15:19-32.
7. Drake C, Nickel C, Burduvali E, Roth T, Jefferson C, Pietro B. The pediatric daytime sleepiness scale (PDSS): Sleep habits and school outcomes in middle-school children. Sleep 2003;26:455-8.
8. Chervin RD, Hedger K, Dillon JE, Pituch KJ. Pediatric sleep questionnaire (PSQ): Validity and reliability of scales for sleep-disordered breathing, snoring, sleepiness, and behavioral problems. Sleep Med 2000;1:21-32.
9. Chervin RD, Weatherly RA, Garetz SL, Ruzicka DL, Giordani BJ, Hodges EK, et al. Pediatric sleep questionnaire: Prediction of sleep apnea and outcomes. Arch Otolaryngol Head Neck Surg 2007;133:216-22.
10. Simard-Tremblay E, Constantin E, Gruber R, Brouillette RT, Shevell M. Sleep in children with cerebral palsy: A review. J Child Neurol 2011;26:1303-10.
11. Sandell DE, O’Brien LM, Shank LK, Warschausky SA. Sleep and quality of life in children with cerebral palsy. Sleep Med 2011;12:252-6.
12. Senanayake N, Román GC. Epidemiology of epilepsy in developing countries. Bull World Health Organ 1993;71:247-58.