



Original Article

Sleep disorders in children with cerebral palsy: neurodevelopmental and behavioral correlates



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ABSTRACT

Objectives: We aimed to estimate the frequency of sleep disorders in children with cerebral palsy (CP) using the Sleep Disturbance Scale for Children (SDSC) and to evaluate the relations between sleep disorders and motor, cognitive, and behavioral problems.

Methods: One hundred and sixty-five children with CP ages 6–16 years (mean age, 11 years) were assessed using the SDSC, the Gross Motor Function Classification System (GMFCS), the Wechsler Intelligence Scale for Children and the Child Behavior Check List (CBCL) to assess sleep, motor, cognitive, and behavioral problems, respectively.

Results: An abnormal total sleep score was found in 19% of children with CP; more than 40% of children had an abnormal score on at least one SDSC factor. The SDSC total score was significantly associated ($P < .01$) with mental retardation, epilepsy, CBCL scores, and level 5 on the GMFCS.

Conclusions: Our results confirm that sleep disorders are common in children with cerebral palsy. The relationship between motor and cognitive behavior and epilepsy should be further explored to better understand how these factors influence one another to identify effective treatments and to improve the well-being of the child.

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1. Introduction

Sleep disorders represent a common problem in infancy and childhood [1–5]. In healthy children, the prevalence of sleep disorders can vary from 5% to 40% [3–5]. This wide range of prevalence is most likely due to the absence of a consensus on what threshold constitutes clinical sleep pathology, the different sample characteristics, the various instruments used to assess sleep, and the duration criteria for defining a sleep problem [6].

Although there are several studies assessing sleep in children with developmental disabilities [6–13], there are only a few in children with cerebral palsy (CP) [6,11–13]; these few studies have reported that sleep disorders are more frequent in CP than in typically developing children. The increased prevalence was related to muscle spasms, musculoskeletal pain, and epilepsy. Newman et al. [6] reported data on 173 children with different types of CP, with 44% of the children presenting with at least one clinically

significant sleep disorder. However, the authors did not consider the presence of mental retardation and behavioral problems, which are known to have a strong influence on sleep [1–9,11,12]. The relationship between sleep and behavior in CP has never been systematically addressed. This issue is particularly relevant, as there is increasing evidence of the high prevalence of behavior problems in children with CP ranging from 26% to 80% [14,15]. Because it is known that sleep disturbances can greatly affect daytime behavior in children leading to neurobehavioral disturbances (e.g., inattention, hyperactivity, learning problems) [1–3,10], we hypothesized that sleep disturbances may be related to daytime behavior in children with CP.

The aims of our study were to estimate the frequency of sleep disorders in children with CP after the age of 6 years and to evaluate the relationship between sleep disorders and motor, cognitive, and behavioral problems.

2. Methods

The children included in our study were part of a collaborative prospective project on families of children with CP regularly

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followed at the Child Neurology Unit of the Catholic University of Rome and the Neurological Institute Besta in Milan, Italy, between January 2010 and December 2012. To have a homogeneous cohort, we only included children with no parental history of a severe or chronic medical condition (e.g., stroke, diabetes mellitus) or a psychologic disorder. The inclusion criteria were children with a diagnosis of CP between the ages of 6 and 16 years with a detailed cognitive and motor assessment. The age range was based on the choice of some assessments performed in the study, for which validation studies and normative data are available from the age of 6 years.

CP was defined as a group of disorders in the development of movement and posture, causing activity limitation attributed to nonprogressive disturbances occurring in the developing fetal or infant brain [16]. Clinical diagnosis was based on the predominant type of motor impairment and was classified according to the criteria proposed by Himmelmann et al. [17]. The children were divided into four groups according to the type of CP: diplegia, hemiplegia, quadriplegia and dyskinesia. All children with CP underwent a comprehensive assessment of motor, cognitive, and behavioral abilities and sleep disturbances. Motor function was evaluated using the Gross Motor Function Classification System (GMFCS) [18] to classify each child's level of gross motor function, with skill levels from 1 to 2 assessing the children's gross motor function by observation.

Cognitive function was measured by using the Wechsler Intelligence Scale for Children III-Revised [19] for children between the ages of 6 and 16 years. The test was performed by a trained psychologist for all the children. Child behavior was assessed using the Child Behavior Check List (CBCL) [20,21]. In this test behavior problems are reported by the child's primary caregiver, the individual who is most responsible for the day-to-day decision making and care of the child. The CBCL consists of 118 items on which parents rate their child's behavior by using 3-point scales: 0 (not true), 1 (somewhat or sometimes true), and 2 (very true or often true). The CBCL provides a total behavior problems score, 2 second-order factor scores (internalizing problems, externalizing problems), and 8 syndrome scores (aggressive behavior, anxious/depressed, attention problems, delinquent behavior, social problems, thought problems, withdrawn, somatic complaints). Raw scores on each clinical factor were transformed to T scores based on published norms. Scores higher than 63 were considered abnormal, scores between 60 and 63 were considered borderline, and scores of less than 60 were considered normal.

Sleep disturbances were assessed using the Sleep Disturbance Scale for Children (SDSC), which showed thorough validation, an adequate level of internal consistency, test-retest reliability, and availability of normative data [3,10]. This scale was originally validated on a sample of 1157 healthy children from the general population aged 6–16 years. It investigates the occurrence of sleep disorders during the previous 6 months and contains 26 items in a Likert-type scale with values from 1 to 5; higher numerical values reflect a higher clinical severity of symptoms. The sum of scores provided a total sleep score with a possible range from 26 to 130. A T score of more than 70 (>95th centile) was regarded as abnormal, and a score of 70 or less was considered to be normal.

The original factor analysis yielded six sleep disturbance factors representing the most common areas of sleep disorders in childhood and adolescence: (1) disorders of initiating and maintaining sleep; (2) sleep-breathing disorders; (3) disorders of arousal, including sleepwalking, sleep terrors, and nightmares; (4) sleep-wake transition disorders (SWTD), including hypnic jerks, rhythmic movement disorders, hypnagogic hallucinations, nocturnal hyperkinesias, and bruxism; (5) disorders of excessive somnolence; and (6) sleep hyperhidrosis (SHY).

This questionnaire was distributed to the primary caregiver of children during the routine neurologic assessment in our units. This questionnaire was completed together with information on the parents' marital status and current parental employment. These demographic data were only used to assess statistical differences between the four CP groups on the requested information. The children also were screened for the presence of epilepsy, which was further categorized into controlled or intractable/active when the seizures were not controlled with treatment, and antiepileptic therapy.

The study protocol was approved by the ethics committee of the institutions and informed consent was obtained from parents.

2.1. Statistical analysis

The children were divided into four groups according to the type of CP. Data were presented as mean values (standard deviations [SDs]) for continuous normally distributed variables, median (interquartile range) for normal continuous variables, and numbers and percentages for categorical variables. The comparisons of the continuous variables, including children's ages, cognitive assessments, scores on SDSC total, and six factors on CBCL scales, were performed using the Kruskal–Wallis equality of populations rank test; the comparisons of the categorical variables (gender, demographic factors, controlled or active epilepsy) and GMFCS scores were performed with the Fisher exact test.

The correlation between SDSC total score and CBCL total scores was explored with Spearman rank order correlation test. Dummy variables for abnormal scores and CBCL scores were created and the association between an abnormal total SDSC score and the physical parameters (i.e., sex, age, CP type, developmental delay, GMFCS level, epilepsy, abnormal CBCL scores) were performed and reported as crude odds ratios (OR) with 95% confidence intervals (CI) and OR adjusted for type of cerebral palsy.

Multivariate analysis was conducted using logistic regression analyses to define the role of specific factors which may affect an abnormal total SDSC score. All of the variables were entered into the initial model. Backward stepwise selection was used to select the variables to enter in the final model with a significance level for the removal and the addition of .3 and .2, respectively. Results of the logistic regression analyses are expressed as OR with 95% CI. A two-tailed value of $P < .05$ was considered significant. Statistical analysis was performed using the Stata Statistical Software: Release 10 (StataCorp LP, College Station, TX).

3. Results

During the study period, 165 children with CP (99 boys; 66 girls) and the primary caregiver fulfilled the inclusion criteria. There were 38 children who presented diplegia (25 boys; 13 girls), 56 presented with hemiplegia (37 boys; 19 girls), 64 presented with quadriplegia (33 boys; 31 girls), and 7 presented with dyskinesia (4 boys; 3 girls). The mean age was 11 years (range, 6–16 years). Sixty-three children were affected by epilepsy (39 with quadriplegia, 17 with hemiplegia, 4 with diplegia, and 3 with dyskinesia) and were all receiving antiepileptic medication. Forty-one out of 63 children showed intractable/active epilepsy (33 quadriplegia, 5 hemiplegia, 2 dyskinesia, 1 diplegia). No statistical difference was found between the mean ages of children in the various forms of CP. No difference was identified in demographic factors or gender (Table 1).

3.1. Cognitive and motor function

Table 2 reports the distribution of motor and cognitive function in children with CP.

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