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Original Article

Sleep disturbances in preschool age children with cerebral palsy: a questionnaire study

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ABSTRACT

Objectives: The study aimed to analyze (i) the prevalence of sleep disorders in pre-school children with cerebral palsy (CP) using the Sleep Disturbance Scale for Children (SDSC), (ii) the possible association with motor, cognitive and behavioral problems, and (iii) the possible differences with typically developing children matched for age and gender.
Methods: One-hundred children with CP (age range: 3–5 years, mean: 3.8 years) were assessed using the SDSC, the Gross Motor Function Classification System (GMFCS), the Wechsler Preschool and Primary Scale of Intelligence, and the Child Behaviour Check List (CBCL) to assess sleep, motor, cognitive, and behavioral problems, respectively. Further 100 healthy children matched for age and sex were assessed using the SDSC.
Results: An abnormal total sleep score was found in 13% of children with CP while 35% had an abnormal score on at least one SDSC factor. SDSC total score was significantly associated with pathological internalizing scores on CBCL and active epilepsy on multivariate analysis. CP group reported higher significant median scores on SDSC total, parasomnias, and difficulty in initiating and maintaining sleep factors.
Conclusions: In pre-school children sleep disorders are more common in children with CP than in healthy control group and are often associated with epilepsy and behavioral problems.

1. Introduction

Several studies in typically developing children have reported that both the type and the prevalence of sleep disorders can vary according to age [1–3]. Infants and toddlers often have difficulties settling to sleep or sleeping through the night. Pre-adolescents show an increased incidence of disorders such as sleep-walking, nightmares, bruxism, and enuresis whereas adolescents are mainly affected by insomnia and daytime sleepiness [1]. A recent review also highlights differences in sleep patterns from the first months of life to adolescence in terms of sleep duration, night waking, sleep latency, longest sleep period, and number of daytime naps [2]. Other authors also report a higher prevalence of dyssomnias and parasomnias in preschool children [3] with a possible correlation with childhood behavior problems, mental retardation or other medical problems [1].

In children with CP, sleep disturbances are more frequent than in typically developing children [4–10], possibly related to motor impairment, pain, behavioral problems or epilepsy. Few studies are available on sleep disturbances in children with CP at different ages. These studies mainly assessed children over 6 years of age and only one included pre-school CP children using questionnaires [10].

The aims of the present study were: (1) to analyze the prevalence of sleep disorders in pre-school children with CP using a structured questionnaire validated for this age; (2) to evaluate differences with typically developing children matched for age and gender; (3) to analyze the relations between sleep disorders and motor, cognitive and behavioral problems.

2. Methods

The children included in this study are part of a collaborative prospective project on families of children with CP regularly followed at the Child Neurology Unit of the Catholic University of Rome and

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at the Neurological Institute Besta in Milan, Italy, between January 2010 and December 2012. The study protocol was approved by the Ethics Committee of the Institutions and informed consent was obtained from parents.

CP was defined as a group of disorders of the development of movement and posture attributed to non-progressive disturbances that occurred in the developing fetal or infant brain, causing activity limitation [11]. Clinical diagnosis was based on the predominant type of motor impairment and classified according to the criteria proposed by Himmelmann et al. [12]. The children were divided into four groups according to the type of CP: diplegia, hemiplegia, quadriplegia, and dyskinesia.

In order to have a homogeneous cohort, we only included children with no parental history of a severe or chronic medical condition (e.g. stroke, diabetes) or psychological disorder. The inclusion criteria were: a diagnosis of CP, age between 3 and 6 years, and a detailed cognitive and motor assessment. A recent study in older children has been published by our group using the same methodological approach [7].

Sleep disturbances were assessed using the Sleep Disturbance Scale for Children (SDSC) validated for pre-school children [13]. It investigates the occurrence of sleep disorders during the previous 6 months, and contains 26 items in a Likert-type scale with values 1–5 (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 as normal.

The factor analysis yielded six sleep disturbance factors representing the most common areas of sleep disorders in pre-school children: parasomnias (PAR) related to arousal disorders (sleepwalking, sleep terrors), nightmares, and sleep–wake transition disorders; difficulty in initiating and maintaining sleep factor (DIMS) related to sleep duration and latency, problems in falling asleep and night awakenings; sleep disordered breathing (SDB); disorders of excessive somnolence (DOES) related to daytime somnolence and sleep attacks; sleep hyperhydrosis (SHY) referred to falling asleep as well as night sweating and nocturnal hyperkinesia; and non-restorative sleep (NRS) concerning items like “the child is unusually difficult to wake up in the morning” or “the child awakes in the morning feeling tired.”

This questionnaire was distributed to the primary caregiver of the children during the routine neurological assessment in our units. Demographic and health questions are not included in the SDSC and were included as a separate partially structured demographic questionnaire. The requested data were gender, age, family status, school education and health status of parents, and age and gender of children. The demographic data were only used to assess statistical differences between the groups on the requested information.

All the children were also screened for the presence of epilepsy, controlled or intractable/active when the seizures fail to come under control with treatment, and antiepileptic therapy.

The SDSC was further distributed to the primary caregivers of a group of children recruited via nurseries and considered as a control group. Questionnaires were filled out by the mothers during the school hours under the supervision of the researchers that distributed the questionnaires (authors: CB, EM, SS); no missing values were reported. All children in the control group attended regular classes in mainstream nursery school and those with obvious or reported sign of mental, developmental or physical disabilities based on school medical records or receiving on-going prescription medication (antiepileptic drugs, antihistaminic drugs, benzodiazepine, melatonin) were excluded.

All children in this study also underwent a comprehensive assessment of motor, cognitive, and behavioral abilities.

Motor function was evaluated using the Gross Motor Function Classification System (GMFCS) [14] to classify each child’s level of gross motor function with skill levels from I to V, assessing the children’s gross motor function by observation.

Cognitive function was measured using the Wechsler Preschool and Primary Scale of Intelligence, 3rd Edition (WPPSI-III) [15] for children between 3 and 6 years of age. The test was performed by a trained psychologist.

Child behavior was assessed using the Child Behaviour Check List (CBCL) [16,17]. In this test behavior problems are reported by the child’s primary caregiver (the person 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, and somatic complaints). Raw scores on each clinical factor were transformed to T-scores based on published norms. Scores >63 are considered abnormal, scores between 60 and 63 as borderline, and scores <60 normal.

2.1. Statistical analysis

Data were presented as mean values (standard deviations [SDs]) for continuous variables normally distributed, median (interquartile range) for non normal continuous data, and count and percentages for categorical variables. The comparisons between the different types of CP for the continuous variables (age of children, cognitive assessments, scores on SDSC total and 6 factors and CBCL scales) were performed using the Kruskal–Wallis equality-of-populations rank test; the comparisons for the categorical variables (gender, epilepsy controlled or active) and GMFCS scores were performed with the Fisher’s exact test.

The comparison between children with CP and control group for SDSC total and the six factor scores and age was performed using the non-parametric Mann–Whitney U test; the comparisons for the gender were performed with the Fisher’s exact test. The association between an abnormal total SDSC score and the physical parameters (sex, age, CP type, developmental delay, GMFCS level, epilepsy, and abnormal CBCL scores) were performed, and reported as crude odds ratios (OR) with their 95% confidence intervals (CI).

Multivariate analysis was conducted by logistic regression to define the role of specific factors that may affect an abnormal total SDSC score. All the significant variables obtained after univariate analysis were entered into the initial model. Results of the logistic regression are expressed as OR with 95% CI.

A two-tailed value of $P < 0.05$ 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, 100 children with CP (52 M, 48 F) and the primary caregiver fulfilled the inclusion criteria: 32 children presented diplegia [14,18], 34 hemiplegia [16,18], 29 quadriplegia [11,19] and 5 dyskinesia [5]. The mean age was 3.8 ± 0.8 years (range 3–5 years). No statistical difference was found among the various types of CP for both mean ages of children (diplegia 3.8 ± 0.8 ys; dyskinesia 4 ± 1 ys; hemiplegia 3.9 ± 0.8 ys; quadriplegia 3.9 ± 0.8 ys) or gender .

Thirty-two children (32%) were affected by epilepsy (18 with quadriplegia, 10 hemiplegia, 4 diplegia) and were all receiving