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Sleep problems and obstructive sleep apnea in children with down syndrome, an overview

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ABSTRACT

Introduction: Children with Down syndrome (DS) have a high prevalence of sleep problems, including behavioural sleep disturbances and obstructive sleep apnea. Sleep problems are associated with a wide range of adverse health effects. Since children with DS are already known to have many comorbidities, they are particularly susceptible for the negative impact of sleep problems.

Aim of this study is (1) to evaluate the prevalence of sleep problems in children with DS, (2) compare the prevalence of sleep problems in children with DS with a community sample of typical developing school-aged children, and (3) to correlate the existence of sleep problems in children with DS and OSA.
Methods: Children enrolled at the multidisciplinary Down team of the University Hospital Antwerp and seen at the ENT department were eligible for this study. The prevalence of sleep problems was evaluated by the use of the Child Sleep Habits Questionnaire (CSHQ) and a full overnight polysomnography was performed to screen for obstructive sleep apnea.
Results: Parents of fifty-four children with DS, aged 7.5 years (5.4–11.6), completed the CSHQ and an overall prevalence of sleep problems was found in 74.1%. In 57.1% of the children OSA was diagnosed with a median obstructive apnea-hypopnea index (oAHI) 7.25/h (5.7–9.8). Overall sleep problems were not age-or gender related, however boys suffer more from daytime sleepiness. Symptoms of sleep disordered breathing correlate with parasomnias, a longer sleep duration and more daytime sleepiness. No correlation was found between sleep problems and underlying OSA.
Conclusion: Children with Down syndrome have a significantly higher prevalence of sleep problems, compared to normal developing healthy school-aged children. We didn't find any correlation between the parental report of sleep problems and underlying OSA, or OSA severity.

1. Introduction

Children with Down syndrome (DS) are at risk to develop sleep problems [1–3]. These include difficulties in initiating and maintaining sleep (DIMS), excessive daytime sleepiness (EDS) and obstructive sleep apnoea (OSA) [4,5]. Overall, sleep problems are not uncommon in school-aged children, however they seem to be definitely more prevalent in children with intellectual disabilities [6,7]. Persistent sleep problems are not benign, and may have a negative impact on health in all its dimensions. Sleep problems have been associated with a wide range of neurocognitive problems, obesity, cardiovascular disease, mood and anxiety disorders, autism spectrum disorders and attention deficit hyperactivity disorder. They may impose a factor of distress for families as well [1]. OSA is highly prevalent in children with DS, with prevalence rates between 24 and 57% in literature [4], compared to 0.7–2% in normal developing children [5]. OSA in children has been associated with cardiovascular problems (pulmonary hypertension, cor pulmonale), obesity, metabolic disturbances, and may affect neurocognition and behaviour. [8]. Children with DS are more susceptible for the negative impact of sleep disturbance and OSA, since these children have frequently pre-existing medical and neurocognitive disabilities. Previous

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studies showed a poor correlation between parental reports of sleep problems and OSA. Therefore, a polysomnography (PSG) has been advocated in every child with DS at the age of 4 years old (or earlier if suspected) [9,10].

Aim of this study is to (1) define the prevalence of sleep problems in children with DS based upon a validated Flemish version of the Child sleep habits questionnaire (CSHQ), (2) compare the prevalence of sleep problems with a community sample of typical developing school-aged children, and (3) to correlate the existence of sleep problems in children with DS and OSA.

2. Methods

2.1. Measurements

2.1.1. Child sleep habits questionnaire

The CSHQ is a 33-item parental questionnaire developed as a screening instrument for sleep problems in school-aged children [11]. Parents are asked to score sleep behaviours over a 'typical' recent week. Every item is scored on a 3-point scale: usually (5 to 7 times/week), sometimes (2 to 4 times/week) or rarely (0 to 1/week). Parents have also the opportunity to indicate if a particular item is perceived as a problem (yes, no or not applicable). The CSHQ allows a total score (>41 in the clinical range, indicating an underlying sleep problem) and 8 subscales or domains: bedtime resistance (6 items), sleep-onset delay (1 item), sleep anxiety (4 items), sleep duration (3 items), night wakening (3 items), parasomnias (7 items), sleep disordered breathing (3 items) and daytime sleepiness (8 items). Three additional questions ask for evening bedtime, morning wake-up and total sleep duration (including daytime sleep). A cut-off score of 41 is believed as the best diagnostic confidence, determined by the intersection point of sensitivity and specificity, respectively 0.80 and 0.72. A cut-off score with maximized sensitivity was sought in previous studies based on the belief that it is more important to avoid false negatives than false positives [11]. The prevalence of the different subscales was evaluated, and a sleep disturbances was considered to be prevalent when at least 20% of the sample reports 'usually' or 'sometimes' [12]. Rare sleep disturbances were defined when less than 5% answered 'usually' or 'sometimes' [12].

In order to gather validated data, the CSHQ was officially translated in Flemish (Linguapolis, language and communication institute, University of Antwerp). In a previous paper, we reported normative data for the CSHQ obtained in a group of healthy Flemish typical developing, school-age children (4–12 years old) [13]. These data are used to compare the prevalence of sleep disorders with the DS children.

The prevalence of sleep problems and the different subscales was compared between children with DS and healthy typical developing school-aged children. Because the age range in the control group is 4–12.9 years old, further analysis is performed with only these children with DS younger than 12.9 years ($n = 44$).

2.1.2. Overnight polysomnography

A full overnight PSG was performed at the Pediatric Sleep Disorders Center of the Antwerp University Hospital, Belgium where the following variables were continuously measured and recorded by a computerized polysomnography (Brain RT, OSG, Rumst, Belgium): electroencephalography (C4/Al and C3/A2); electro-oculography; electromyography of anterior tibialis and chin muscles; and electrocardiography. Respiratory effort was measured by respiratory inductance plethysmography and oxygen saturation by a finger probe connected to a pulse oximeter. Airflow was measured by means of nasal pressure cannula and thermistor, and snoring was detected by means of a microphone at the suprasternal notch. Children were also monitored on audio/videotape using an infrared camera. Polysomnograms were manually scored by certified technicians according to international guidelines [14]. A diagnosis of OSA is established with and obstructive apnea/hypopnea index (oAHI) $\geq$ 2/h [15].

2.2. Study population

All children with DS (aged 4–18 years) enrolled at the multidisciplinary Down team (University Hospital Antwerp, Belgium) and seen at the ENT department were eligible for inclusion. Children were divided in 4 age groups: age group 1 (4–6.9 years), age group 2 (7–8.9 years), age group 3 (9–10.9 years) and age group 4 (>11years). Parents completed the CSHQ on the day of consultation at the ENT department or at home.

The study was approved by the local Ethical Committee (B300201213390), and all parents gave written informed consent before inclusion in this study.

2.3. Data analysis

All statistical analyses were performed using the Statistical Package for the Social Science version 20.0 (SPSS 20.0). Data were explored for normality using the Kolmogorov Smirnov test. Data are presented as the median with lower and upper quartile. Correlation analysis was performed with a linear regression analysis (Spearman correlation), corrected for age and gender. Group differences were evaluated with a Mann Whitney U test. Group differences for categorical variables were tested with Chi-square test. A p-value of <0.05 was considered as statistically significant.

3. Results

The parents of fifty-four children with DS completed the CSHQ. Median age was 7.5 (5.4–11.6) years, and most children ($n = 33$, 61%) were included in the younger age groups (age group 1 ($n = 22$), age group 2 ($n = 11$), age group 3 ($n = 4$), age group 4 ($n = 17$)). Fifty-five percent of the children were male and the median CSHQ score in our study population was 44.0 (40.0–48.0). Polysomnography was performed in 42 of these children (77.8%). We found an overall prevalence of sleep problems (CSHQ in the clinical range) in 74.1% of the studied population, with a median score 46.0 (44.0–50.8). In 57.1% of the children undergoing PSG, a diagnosis of OSA was established with median oAHI 7.25/h (5.7–9.8).

In this study, overall sleep problems (CSHQ score in the clinical range) were not age–or gender related. However parasomnias and sleep onset delay appeared to be age related. Parasomnias were negatively associated with age ($p = 0.014$ ($B = -0.333$)). If the youngest age group (4–6.9 years) was compared to the oldest age group (>11 years), a highly significant difference was found ($p = 0.007$). Also sleep onset delay was significantly more prevalent with increasing age ($p = 0.046$ ($B = 0.273$)). Boys suffer more from daytime sleepiness, with median scores in the subscale of daytime sleepiness of 13 (10–15) compared to 10 (9–12) for girls ($p = 0.024$ ($B = 0.306$)). Median age of these boys was 8.2 years (6.0–12.1).

Children with sleep anxiety have a significant longer sleep onset delay ($p = 0.007$, $B = 0.721$). These children have also a higher bedtime resistance ($p = 0.000$, $B = 0.388$), and tend to sleep longer ($p = 0.060$). Symptoms of sleep-disordered breathing (SDB) are significantly correlated with parasomnias ($p = 0.002$, $B = 0.405$), longer sleep duration ($p = 0.022$, $B = 0.322$) and more daytime sleepiness ($p = 0.009$, $B = 0.364$). Children who awake frequently during the night sleep significant longer ($p = 0.010$, $B = 0.362$).

The most prevalent sleep disturbances are summarized in Table 1. In 59.3% of the children a restless sleep was reported, and