Predicting the presence of sleep-disordered breathing in children with Down syndrome

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ABSTRACT

Objective: Sleep-disordered breathing (SDB) is highly prevalent in children with Down syndrome. Given the scarcity of resources and the presence of risk factors for SDB in this population, the objective of this study is to identify the clinical predictors of SDB, which would assist prioritization of children with Down syndrome for SDB evaluation.

Methods: A retrospective cohort study was conducted on children enrolled in the Down syndrome clinic at CHEO who underwent polysomnography in 2004–2014. Total apnea-hypopnea index (AHI) or obstructive AHI (OAHI) > 5 events/hour was considered clinically significant. Associations between SDB and concurrent diagnoses, referral reasons, and sleep symptoms assessed by questionnaire were examined using Pearson’s chi-square test or Fisher’s exact test as appropriate. Univariate and multivariate logistic regression analyses were used to examine the predictors of SDB.

Results: SDB was present in 42.9% of 119 children, with its highest prevalence at age 8 years. Symptoms were not significantly associated with AHI > 5 events/hour or OAHI > 5 events/hour. Gastroesophageal reflux was associated with lower odds of OAHI > 5 events/hour on univariate testing (odds ratio 0.16, 95% CI 0.04–0.72; p = 0.02) and multivariate analysis (odds ratio 0.05, 95% CI 0.0006–0.50; p = 0.002).

Conclusions: SDB is highly prevalent at all ages in children with Down syndrome. Symptoms did not predict SDB in this population, although gastroesophageal reflux may mimic SDB, which indicates that clinicians should continue to perform ongoing surveillance for SDB throughout the lifespan of children with Down syndrome.

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1. Introduction

Sleep-disordered breathing (SDB) impairs gas exchange during sleep, related to airway obstruction (obstructive sleep apnea), abnormal respiratory drive (central sleep apnea) or effort, or a mix of both conditions [1]. The prevalence of SDB in children with Down syndrome is 50% (30–60%) [2] compared to 1–5% in the general population [2]. Children are at an increased risk of SDB because of their small oropharynx, narrow upper airway structure, mid-facial hypoplasia, relatively large tongues, and low tone [3], which may be compounded by adenoidal and tonsillar hypertrophy. SDB can lead to cardiac complications, such as pulmonary and systemic hypertension [4], and insulin resistance [5,6]. Because of the significant comorbidity associated with Down syndrome and SDB, the identification of key clinical symptoms that predict SDB is critical to facilitate early diagnosis and treatment.

The gold standard test to diagnose the presence and the severity of SDB is a polysomnography (PSG) [7] because physical exams and medical histories are not sensitive and specific enough to confirm SDB in the general population [8]. Clinical care guidelines for children with Down syndrome recommend that a PSG is performed by aged four years [9,10]. There is, however, insufficient capacity to meet this requirement, as PSGs are usually performed at tertiary institutions.
care children’s hospitals and have excessively long waitlists of up to two years [11]. To prioritize children for PSG evaluation, distinguishing clinical features that predict SDB in children with Down syndrome need to be identified.

The identification of clinical predictors of SDB in the Down syndrome population will assist with more effective prioritization of these children for PSG, which will lead to a more rapid identification and treatment of SDB in this population. This study aimed to determine, among children with Down syndrome who have sleep symptoms on screening history, which symptoms are most predictive of SDB on PSG.

2. Methods

2.1. Study population and ethics

A retrospective cohort study was conducted of children enrolled in the Down syndrome clinic at the Children’s Hospital of Eastern Ontario (CHEO) who underwent a PSG between 2004 and 2014. This research project was approved by the CHEO Research Ethics Board.

2.2. Polysomnographic evaluation

The Down syndrome clinic at the CHEO follows all children with Down syndrome within the catchment area of the hospital at regular intervals. At each clinic visit, children are screened for sleep problems and if these are present, the children are then referred for polysomnographic evaluation. Only the first PSG performed for each child is included in the present study.

PSGs were performed and scored according to the American Association of Sleep Medicine pediatric standards by using Harmonie-E Version 7.0a from 2004 to 2010 or Neuroworks Database Version 8.0 from 2010 to 2014 (Natus Medical Incorporated, Pleasanton, CA, USA) [12]. Each PSG was scored using the guidelines of the American Academy of Sleep Medicine that were in effect at the time the study was conducted. This included the monitoring of four electroencephalogram leads, electro-oculogram, submental and tibial electromyogram, in addition to chest and abdominal wall inductance plethysmography, airflow measurements (nasal pressure), oxygen saturation, transthecal and end-tidal carbon dioxide measurements, and video and audio recordings. Studies were interpreted and categorized by one of two pediatric sleep medicine physicians as SDB present or absent. If present, SDB was classified as obstructive or central. Mixed events were included in the obstructive index [13]. The severity of SDB was categorized using the apnea hypopnea index (AHI). A total AHI > 5 events/hour was considered clinically significant SDB as reported in other studies [12]. In this study, the types of SDB were distinguished as follows. An obstructive mixed AHI (OAHI) > 5 events/hour was considered clinically significant SDB as reported in other studies [2]. In this study, the types of SDB were distinguished as follows. An obstructive mixed AHI (OAHI) > 5 events/hour was considered clinically significant obstructive sleep apnea. Central sleep apnea was considered to be present if the total AHI was >5 events/hour, but the OAHI was <5 events/hour and the majority of respiratory events were central. AHI > 5 and OAHI > 5 were considered co-primary outcomes. Hypoventilation was defined according to the American Academy of Sleep Medicine criteria of CO2 above 50 mmHg for at least 25% of total sleep time [12]. Finally, reasons for referral indicated by the referring physician on the PSG requisition were explored. These included witnessed apneas, daytime sleepiness, restless sleep, behavioral issues, pulmonary hypertension, snoring, or other.

2.3. Sleep study questionnaire

During the night of each PSG, a sleep study questionnaire based on the pediatric sleep questionnaire [14] was completed by parents/guardians. This questionnaire included information on the following symptoms: snoring, gasping for air, witnessed apneas, struggling to breathe during sleep, choking sounds, awakenings during the night, restless sleep, and appearing congested. The frequency of symptoms was categorized as never, rarely, sometimes, or often.

2.4. Medical records

Data for this study were additionally collected from medical records to obtain information on concurrent diagnoses including asthma/respiratory conditions, repaired congenital heart disease, current cardiac issues, gastroesophageal reflux disease (GERD)/aspiration/GI issues, obesity, behavioral issues/anxiety/ADHD, neurological issues, adenotonsillar hypertrophy, diabetes/metabolic syndrome, failure to thrive, rhinitis/rhinorrhea/sinusitis, or scoliosis. Height and weight values were also obtained from the medical record, from which BMI z-scores were calculated using growth charts for children with Down syndrome [15].

2.5. Statistical analysis

Characteristics of study participants were summarized descriptively. Categorical variables were described using frequency and percentage. Continuous variables were summarized using mean, standard deviation, median, interquartile range, and range, as appropriate. The relationship between the prevalence of SDB and the age at first PSG was examined by logistic regression. Additionally, corresponding to current recommendations that PSG be performed by age four in this population, Pearson’s chi-square test was used to examine the association between each of OAHI > 5 or AHI > 5 and age at PSG dichotomized between under four years and four years or older. Associations between AHI > 5 and OAHI > 5 and concurrent diagnoses, as well as SDB and sleep questionnaire responses (dichotomized between always/sometimes vs. rarely/never) were examined using Pearson’s chi-square test or Fisher’s exact test as appropriate. These were summarized using odds ratios (OR) and 95% confidence intervals. Two-sided p-values <0.05 were deemed statistically significant. Similar analyses were conducted for AHI > 5. Predictors of AHI > 5 (concurrent diagnoses and/or sleep study questionnaire items) with p-values <0.10 were included in a multivariate logistic regression. A similar model was constructed for predictors of OAHI > 5. The variance inflation factor (VIF) was used to assess for multicollinearity. Given the limited sample size of patients with SDB, no interactions were modeled. Because two primary outcomes were examined, a Bonferroni correction for multiple testing was applied to the results of the multivariate analysis. All analyses were performed using SPSS version 23 [16].

3. Results

3.1. Demographics

A total of 506 children were followed at the CHEO Down syndrome clinic between 2004 and 2014. Of these, 122 children underwent PSG. Three PSG had incomplete or inconclusive data for interpretation. A total of 119 children who underwent PSG were included in the analysis for this study (Table 1). Their age ranged from 2.5 weeks to 16.8 years.

3.2. Reasons for referral

Snoring was the most frequent reason for referral in 67% of cases, while apnea was the reason for referral in 27% of cases.
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