International Journal of Pediatric Otorhinolaryngology 83 (2016) 47–50

Contents lists available at ScienceDirect

International Journal of Pediatric Otorhinolaryngology

journal homepage: www.elsevier.com/locate/ijporl

Sleep-disordered breathing in children with Down syndrome: Usefulness of home polysomnography

Pablo E. Brockmann [a,b,*], Felipe Damiani [c], Felipe Nuñez [c], Ana Moya [a], Eduardo Pincheira [b], Maria A. Paul [d], Macarena Lizama [e]

[a] Department of Pediatric Cardiology and Pulmonology, Division of Pediatrics, School of Medicine, Pontificia Universidad Católica de Chile, Santiago, Chile
[b] Sleep Medicine Center, Department of Neurology, School of Medicine, Pontificia Universidad Católica de Chile, Santiago, Chile
[c] School of Medicine, Pontificia Universidad Católica de Chile, Santiago, Chile
[d] Division of Pediatrics, School of Medicine, Pontificia Universidad Catolica de Chile, Santigo, Chile
[e] Down Syndrome Center, Pontificia Universidad Católica de Chile, Santiago, Chile

ARTICLE INFO

Article history:
Received 19 October 2015
Received in revised form 20 January 2016
Accepted 23 January 2016
Available online 1 February 2016

Keywords:
Snoring
Apnea
Diagnostic
Down syndrome
Trisomy 21
Polysomnography

ABSTRACT

Objective: To investigate the technical feasibility of unattended home polysomnography (HPSG) in children with Down syndrome.
Methods: Data from children with Down syndrome under 10 years of age referred to a diagnostic sleep study was analyzed. A full sleep-lab based polysomnography (PSG) or a HPSG with a portable device was performed. Uninterpretable HPSGs were defined as: recordings with (i) loss of ≥2 of the following channels: nasal flow, or thoracoabdominal sensors, or (ii) HPSG with less than 4 h of artifact-free recording time or (iii) less than 4 h SpO_2 (peripheral capillary oxygen saturation) signal.
Results: A total of 44 children (68% males) were included in the study, with a mean age of 3.6 (0.1–10) years. PSG was performed in 8 cases and HPSG in 36 cases. Six HPSG recordings were classified as uninterpretable and had to be repeated. Age, gender and BMI were no significant predictors of uninterpretability of the HPSG. Obstructive sleep apnea (OSA) was present in 61% ($n = 27$) of all subjects, and classified as mild, moderate, and severe in 43% ($n = 19$), 11% ($n = 5$), and 7% ($n = 3$) of cases, respectively. Interpretable and technically acceptable HPSGs were obtained in 30 subjects (83%). Age, gender and BMI were no significant predictors for interpretability of the HPSG.
Discussion: This study demonstrates that a portable polysomnographic home device may be helpful for diagnosing OSA in children with Down syndrome. Considering the potential consequences of untreated OSA, this screening test may be helpful for early diagnosis of OSA in children with Down syndrome.

1. Introduction

Sleep-disordered breathing (SDB) has a high prevalence among children with Down syndrome. The spectrum of SDB that affects this group of children ranges from primary snoring to obstructive sleep apnea (OSA), the latter with a reported prevalence of 31% to 63% [1,2], which accounts for a 10- to 20 fold higher prevalence of OSA than that observed in children without Down syndrome [3].

Children with Down syndrome have many several predisposing factors for SDB, including midface hypoplasia, mandibular hypoplasia, glossoptosis, a small upper airway, and enlarged tonsils [4]. In addition, children with Down syndrome have a small and hypotonic airway and an increased incidence of lower respiratory tract anomalies [5,6].

The consequences of untreated OSA may result in serious problems including poor academic performance [7,8], behavioral problems [9], hyperactivity [10], attention difficulties [11], and worsening of mental function [10]. Considering the growing body of evidence that links OSA with neurocognitive issues in children [10], the impact of untreated OSA on the mental function in children with Down syndrome is extremely concerning.

The high prevalence and serious consequences of OSA in children with Down syndrome have led to recommendations for screening all children with this condition for OSA at 5 years of age [12]. The presence of snoring, disturbed sleep, awakenings, and daytime symptoms like somnolence or hyperactivity may lead to earlier consultation and treatment. However, parents of children with Down syndrome may underestimate the severity of the sleep

* Corresponding author at: Department of Pediatric Cardiology and Pulmonology, Department of Pediatrics, School of Medicine, Pontificia Universidad Católica de Chile, Lira 85 5to piso, 8330074 Santiago, Chile. Tel.: +56 2 23543767.
E-mail address: pbrockmann@med.puc.cl (P.E. Brockmann).

http://dx.doi.org/10.1016/j.ijporl.2016.01.030
0165-5876/

disturbances and overlook the presence of OSA [13]. Furthermore, OSA cannot be diagnosed based solely on clinical history or physical examination [7], a full-night sleep lab-based polysomnography (PSG) is currently the recommended gold standard for diagnosis [8]. Nevertheless, this test may be especially difficult to perform in children with Down syndrome, considering it involves an overnight hospital stay, away from the regular surroundings of the child. A noninvasive, home-based study for diagnosing OSA seems to be therefore an interesting solution. Among the few studies that have demonstrated adequate diagnostic accuracy and feasibility [14], unattended portable polysomnography (HPSG) has shown promising results [10].

The feasibility of HPSG in a pediatric setting has been previously demonstrated [9,14], however, this has not been documented in children with Down syndrome. Therefore, the aim of the present study was to investigate the technical feasibility of unattended HPSG using portable equipment in children with Down syndrome. In addition, HPSG and PSG results were compared.

2. Methods

2.1. Subjects

We included all data from children with Down syndrome aged under 10 years who were referred to a sleep study between 2013 to 2015 at the Sleep Laboratory or the Respiratory Laboratory of the Pontificia Universidad Católica de Chile, Santiago, Chile. Only data of children with confirmed Down syndrome were selected for this study. These children were sent to perform either a HPSG, or PSG based on the clinical judgment of their physicians. The main reason for requesting the sleep study was habitual snoring, defined as snoring for more than 3 nights per week. No prior screening tool was used for the selection.

Demographic data, nutritional status, and health records were obtained and registered into the dataset. Nutritional status was assessed using body mass index (BMI, kg/m^2). Age and gender-specific z-scores were obtained for each subject's BMI. Associated autism spectrum disorder was recorded if a pediatric neurologist had made the diagnosis. Other associated comorbidities such as congenital heart disease, hypothyroidism, respiratory problems, and use of supplemental oxygen or mechanical ventilation were obtained from the subject's health records. The study and use of the subject's clinical data was approved by the Ethics Committee of the Faculty of Medicine at the Pontificia Universidad Católica in Santiago, Chile (Approval number 14-327).

2.2. Procedures

For HPSG, a portable cardiorespiratory device was used unattended at home (Embletta® Gold III, Embla, Broomfield, Colorado, USA). This procedure has been previously reported by our group in habitually snoring children [14]. The following six channels were recorded: (i) nasal flow using a pressure transducer cannula, (ii) thoracic movements, (iii) abdominal movements, (iv) pulse oximetry, (v) heart rate measured by electrocardiography, (vi) position sensor. The device used for the HPSG was installed in the respiratory lab and returned by the parents of the children the next morning. A hotline number was given to parents in order to answer questions or solve problems.

Those patients sent for PSG used a computerized polysomnographic system (ALICE 5.0, Respironics, Andover, MA, USA). PSG was performed in the sleep lab at night with continuous attendance. The study montage included the following channels: 3-lead electroencephalography, 2-lead electrooculography, 3-lead submentalis electromyography, chest and abdominal wall movements, nasal pressure transducer, snoring, pulse oximetry-derived arterial hemoglobin oxygen saturation and pulse waveform, heart rate, digital audio and video.

For both PSG and HPSG, respiratory events and sleep architecture were analyzed according to current criteria [15]. Arousals were identified as defined by the American Sleep Disorders Association Task Force report [16]. Central, obstructive, and mixed apneas and hypopneas were identified according to current recommendations [17]. Obstructive apneas were defined as the absence of airflow with continued chest wall and abdominal movement for the duration of at least two breaths. Central apneas were defined as the absence of nasal flow and thoraco-abdominal movements for more than 20 s, or for more than 2 breaths if the episode was accompanied by desaturation or arousal. Hypopneas were defined as a decrease in nasal flow of at least 30% with a corresponding decrease in SpO_2 (peripheral capillary oxygen saturation) of 3% or more and/or an arousal [17]. The apnea–hypopnea index (AHI) was calculated based on the number of obstructive and mixed apneas and hypopneas per hour of total sleep time. OSA was defined as an AHI >1 [17]. Mild, moderate, and severe OSA was defined as an AHI <5, >5, and >15, respectively. All studies were reported by the same investigator.

2.3. Evaluation of feasibility

Based on the feasibility criteria of our previous publication on habitually snoring children [14] we determined the need for a new recording due to uninterpretability as the main failure criteria. An uninterpretable study was defined as those with $\geq$ of the following criteria: (i) loss of the following channels: nasal flow, or thoracic or abdominal sensors, (ii) recordings with less than 4 h of artifact-free recording time or (iii) less than 4 h of SpO_2 signal [14]. These criteria were applied to both PSG and HPSG.

2.4. Statistics

Descriptive statistics were used to summarize the children's demographic and polysomnographic characteristics (i.e., numbers, percentages, median, minimum, maximum for non-normal distributed data; and mean and standard deviation for data with normal distribution). Comparisons between PSG and HPSG were conducted using Mann Whitney *U*-Test for not-normal data, and Student's *t*-test for normally distributed variables. Factors that may have influenced the interpretability of the recordings were investigated using logistic regression. Age, gender, AHI, and BMI z-scores were analyzed as independent variables in the logistic regression equation. Odds ratios and their 95% confidence intervals (95% CI) were calculated. Statistical software SPSS 20.0 (Statistical Package for the Social Science 20.0 for Mac) was used for all analyses. A p-value <0.05 was considered statistically significant.

3. Results

Of the $n = 44$ children included in the study, $n = 30$ (68%) were males. Mean age was 3.6 (0.1–10) years. A total of $n = 8$ PSG, and $n = 36$ HPSG were performed in the patients. A diagnosis of congenital heart disease was present in 27 (61%) cases, autism spectrum disorder in $n = 4$ (9%) (all of them male, aged 6, 4, 3, and 3 y), hypothyroidism in $n = 27$ (61%), and swallowing/feeding disorder in $n = 13$ (30%) cases. Table 1 shows the demographic and clinical characteristics of the HPSG and PSG groups, none of them were statistically significant, except for the use of home oxygen in $n = 3$ (43%) versus $n = 2$ (6%) children, respectively ($p = 0.024$).

OSA was diagnosed in $n = 27$ (61%) of all subjects. OSA was classified as mild, moderate, and severe in $n = 19$ (43%), $n = 5$ (11%), and $n = 3$ (7%) children, respectively. There were $n = 7$ (88%) children with OSA in the PSG group, compared to $n = 20$ (56%) in