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Clinical variables determining the success of adenotonsillectomy in children with Down syndromed

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ABSTRACT

Objectives: To evaluate the evolution of polysomnographic parameters of children with Down syndrome and obstructive sleep apnea syndrome submitted to adenotonsillectomy and the interaction of comorbidities on therapeutic outcome.
Methods: Ninety patients with Down syndrome and habitual snoring were identified between 2005 and 2015 in a Pediatric Otorhinolaryngology Clinic. Parent's complaints were evaluated by the test of equality of two proportions. Wilcoxon test was used to examine pre- and post-operative polysomnographic differences. Mann-Whitney test evaluated the influence of comorbidities. A $p < 0.05$ was considered significant.
Results: A total of 27 patients met the inclusion criteria (55.6% patients were males; mean (SD) age were 6.7 (3.6) years (range, 1.5–16 years). Significant improvement of parent's complaints ($p < 0.001$), arousal index ($p = 0.045$), and minimum oxygen saturation were observed post-adenotonsillectomy ($p = 0.034$). Adenotonsillectomy was able to resolve obstructive sleep apnea syndrome in 29.6% of children with Down syndrome. Nineteen patients (70.4%) remained with obstructive sleep apnea syndrome and 44.4% showed a reduction of at least 50% of obstructive apnea-hypopnea index. Central apnea index post-adenotonsillectomy was worse in patients with heart disease ($p = 0.022$). Sleep efficiency ($p = 0.031$), N1 sleep stage ($p = 0.001$), apnea-hypopnea index ($p = 0.023$), and central apnea index ($p = 0.008$) were worse after surgery in patients with hypothyroidism. Patients with severe OSAS showed significant improvement in polysomnographic parameters after surgery.
Conclusion: Although adenotonsillectomy improved symptoms and objective sleep data in children with Down syndrome, it was not able to resolve obstructive sleep apnea syndrome in most patients. Congenital heart diseases and hypothyroidism may affect the outcome.

1. Introduction

Down syndrome (DS) is the most common chromosomal disorder, with an estimated incidence of 1: 1000 live births worldwide and 1.13: 1000 live births in Brazil [1–3].

Obstructive sleep apnea syndrome (OSAS) may prevent these patients from reaching their full developmental capacity. The prevalence of OSAS in children with Down syndrome has been estimated at 57%–66.4% in studies that evaluated data obtained from full-night polysomnography assessment [4–6].

The possible causes for the high prevalence of OSAS in these patients with DS are the craniofacial anatomical alterations found in this specific population, such as middle-third facial hypoplasia, micrognathia, flaccid supraglottis, narrow nasal fossae and oropharynx [7,8]. Hypertrophy of the pharyngeal and palatine tonsils, reduced-volume oral cavity, narrow palate, glossoptosis, relative macroglossia, increased secretion production, cervical fat

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deposition, hypothyroidism and generalized muscular hypotonia are also contributing factors [7,9–11].

In healthy children, OSAS results in growth deficit, developmental impairment, behavioral problems, low academic performance, systemic and pulmonary arterial hypertension. In children with DS, OSAS also impairs executive function and is associated with poor verbal fluency and lack of inhibition, which may hinder the ability of these patients to function independently [9,10,12].

Polysomnography (PSG) is the gold standard for the diagnosis of sleep-disordered breathing and it helps therapeutic decision-making and surgical indication [13]. PSG prior to adenotonsillectomy (AT) in DS patients is crucial, as it defines OSAS severity and predicts the need for postoperative monitoring. Postoperatively, the PSG is also indicated in these patients, as it identifies children with residual OSAS who may require further treatment [13,14]. Although it is not curative in all patients with DS, the adenotonsillectomy (AT) is the treatment of choice for OSAS in these patients [7].

The aim of this study was to evaluate the evolution of clinical complaints and polysomnographic parameters in children with OSAS and DS submitted to adenotonsillectomy. We also planned to verify the interaction of comorbidities (heart disease and hypothyroidism) in the therapeutic outcome.

2. Material and methods

A longitudinal study of a historical cohort of children with DS and habitual snoring, treated at the Pediatric Otorhinolaryngology Outpatient Clinic of Universidade Federal de São Paulo, São Paulo, SP, Brazil, was carried out from 2005 to 2015. The patient's files were identified in a database based on the diagnosis according to the International Classification of Diseases (ICD 10) codes Q90.9 for DS, G47.3 for sleep apnea and J35.3 for pharyngeal and palatine tonsil hypertrophy. The polysomnography assessments were performed at Instituto do Sono, Department of Psychobiology, Universidade Federal de São Paulo.

The legal guardians of the children selected for the study were invited to return to the clinic by telephone contact. They were informed about the study and signed the Informed Consent. The research project was approved by the Institutional Research Ethics Committee under number 41620415.1.0000.5505.

The inclusion criteria were the diagnosis of OSAS, defined by the obstructive apnea-hypopnea index (O-AHI) $\geq$ 2.0 events per hour and a minimum percutaneous oxygen saturation value < 92% [15], the indication of surgical intervention, represented mainly by AT, followed by postoperative PSG.

We excluded patient who lack pre- or post-operative PSG, or who had upper airway comorbidities (subglottic stenosis, laryngomalacia), and sleep period <180 min.

The complaints reported by the parents were compared before and after the surgery using the test of equality of two proportions. Polysomnographic parameters pre- and post-adenotonsillectomy were analyzed using Wilcoxon test. The Mann-Whitney test was used to evaluate the influence of comorbidities. The significance level was set at 5% for this study.

Nutritional status was classified in percentiles according to their distribution in the weight-for-age growth curves for children with DS proposed by Cronck [16].

The PSG was carried out at night in a dark and silent room, with ambient air and in the company of the child's parent or tutor, without sleep deprivation or sedation. The electrophysiological and cardiorespiratory parameters were recorded in a computerized system (Embla-N7000, Embla Systems, Inc., Broomfield, CO, USA): electroencephalogram (F4/M1, F3/M2, C4/M1, C3/M2, O2/M1, O1/M2), bilateral submental and tibial electromyography, right and left electrooculogram, airflow through nasal pressure cannula and oronasal thermistor, thoracic and abdominal respiratory effort by uncalibrated inductance plethysmography, percutaneous oxygen saturation by pulse oximetry, snore sensor (microphone), and body position. Sleep staging, body movements and respiratory events were evaluated according to the American Academy of Sleep Medicine criteria [17]. Obstructive apnea was staged when there was absence of oral and nasal airflow in the presence of thoracoabdominal movements lasting more than or equal to 2 respiratory cycles. Hypopnea was defined as a reduction of at least 50% in the oronasal flow amplitude with duration greater than or equal to 2 respiratory cycles, associated with a reduction greater than or equal to 3% in oxygen saturation and/or an arousal.

OSAS was defined when there was obstructive apnea and hypopnea index (O-AHI) >2.0 events per hour [15] and a minimum percutaneous oxygen saturation value < 92%. Patients with O-AHI between 2 and 4.9 were classified as having mild OSAS, whereas those with O-AHI between 5 and 9.9 were classified as moderate, and those with O-AHI $\geq$ 10 as severe OSAS. Patients with O-AHI <2.0 and oxygen saturation value > 92% were classified as having primary snoring.

The primary endpoint measures were patient evolution changes in the PSG parameters after surgery and the secondary endpoints were O-AHI <2.0 and a decrease of at least 50% in O-AHI.

To calculate the sample size, the frequency of OSAS resolution of 16.6% was considered as the primary outcome. This value represents the percentage of children who achieved complete resolution of OSAS, defined as O-AHI <1 in a previous study [18]. A confidence interval of 95% and a nominal error of 5% were taken. Based on these values, the ideal sample would consist of 43 patients.

3. Results

Ninety patients with DS and habitual snoring were identified. Sixty-three patients were excluded: 23 were on surgical planning, 25 were not submitted to preoperative or postoperative PSG, 3 had total sleep time <180 min, and 12 had primary snoring.

The characteristics of the final sample, consisting of 27 patients with OSAS, can be seen in Table 1. The mean age at the date of surgery was 6.7 ± 3.6 and ranged from 1.5 to 16 years. Data of excluded patients were similar to data of patients included in the study.

Twenty-six children were submitted to AT and one to adenoidectomy alone. In nine (33%), the immediate postoperative period was carried out in an intensive care unit. Complications occurred in three cases: one child had a cardiorespiratory arrest during extubation, which was promptly reversed. Hoarseness, requiring nebulized epinephrine, was detected in two other children.

The parents of patients with OSAS were questioned regarding

Table 1
Patient characteristics.

	OSAS (27)	Excluded (63)
Age (years)	6.7 ± 3.6	6.3 ± 3.3
Male gender	15 (55.6%)	37 (58.7%)
Weight (kg)	23.4 ± 12.6	20.1 ± 10.2
< P5	2 (7.4%)	1 (2.3%)
P5-P25	1 (3.7%)	7 (15.9%)
P25-P75	17 (63.0%)	21 (47.7%)
P75-P95	4 (14.8%)	11 (25.0%)
≥ P95	3 (11.1%)	4 (9.1%)

Quantitative data expressed as mean ± standard deviation and qualitative data expressed as number and proportion N (%). Distribution according to the weight-for-age chart in percentiles (P5 to P95). OSAS: obstructive sleep apnea syndrome. Excluded: data from excluded patients.