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Original Article

Drug-induced sedation endoscopy in surgically naive children with Down syndrome and obstructive sleep apnea

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ABSTRACT

Objective: To describe the pattern of upper airway (UA) obstruction in surgically naive children with Down syndrome and obstructive sleep apnea (OSA), and to evaluate the outcome of drug-induced sedation endoscopy (DISE)-directed treatment.
Methods: A prospective study of DISE in surgically naive children with Down syndrome and OSA was performed. Treatment was individually tailored based on the DISE findings and was evaluated by control polysomnography (PGS). Results are presented as median (lower–upper quartile) unless otherwise stated.
Results: In 41 children, aged 4.2 years (range, 2.8–6.0) with a body mass *z* score of 1.04 (–0.55 to 1.82) and obstructive apnea–hypopnea index (oAHI) of 10.1/h (range, 6.3–23.0), DISE was performed. Adeno-/tonsillar obstruction was found in 75.6% of the patients, and these patients subsequently underwent UA surgery. Seven patients were non-surgically treated, and three received a combined treatment. A multilevel collapse was present in 85.4%. Tongue base obstruction was present in ten patients (24.4%) and epiglottic collapse in 48.8%. Pre- and postoperative PSG data were available for 25 children (adenotonsillectomy, n = 16; tonsillectomy, n = 7; adenoidectomy, n = 2). A significant improvement in oAHI from 11.4/h (range, 7.7–27.0) to 5.5/h (range, 2.1–7.6) was found. Persistent OSA was present in 52% of the children. No significant association between different DISE findings and persistent OSA could be found.
Conclusion: Most patients with Down syndrome and OSA present with multilevel collapse on DISE. Adenotonsillectomy results in a significant improvement of the oAHI; however more than half of the patients had persistent OSA, probably due to multilevel collapse. Upper airway evaluation may provide more insights into the pattern of UA obstruction in patients with persistent OSA.

1. Introduction

Children with Down syndrome (DS) are predisposed to a number of health problems affecting their development and quality of life, as described in several earlier reports [1–3]. The prevalence of obstructive sleep apnea (OSA) in children with DS is 30–60%, which is substantially higher compared to 1–4% in a general pediatric population [3]. Multiple factors predispose children with DS to upper airway (UA) obstruction, such as midfacial hypoplasia, relative macroglossia (due to smaller bony framework of mandible and maxilla), hypotonia, obesity, hypothyroidism, an immature immune system with more respiratory infections, and a higher risk of gastroesophageal reflux. Furthermore, laryngomalacia, subglottic stenosis, and tracheomalacia are more frequently reported in children with DS [4,5].

All of these factors contribute to the high risk of developing complex UA obstruction in children with DS. Adenotonsillectomy (AT) is the first-line treatment for pediatric OSA, with a success rate of 71–87% in a normally developing pediatric population [6,7]. The success rate of AT in children with DS and OSA is, however, poor and less favorable compared to that in normally developing children. This has been attributed to multilevel UA obstruction in children with DS [8,9]. Persistent OSA has been observed in 30–50% of children with DS following adenotonsillectomy [8,10].

A thorough and detailed evaluation of the UA should be performed to identify the level and degree of UA obstruction in children known to have multilevel UA collapse, such as those with DS. Depending on these results, an individually tailored treatment may be proposed to optimize treatment outcome. Different techniques are reported in the literature to evaluate the UA in children with OSA. A detailed clinical examination should always be performed, and awake flexible laryngoscopy with or without imaging can be

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done, although neither allows a dynamic evaluation of the airway during sleep. Cine-magnetic resonance imaging (MRI) may provide a high-resolution examination of the dynamic airway, particularly in children with multilevel obstruction [11]. However, experience with this technique is limited mainly to a single center [12]. Drug-induced sedation endoscopy (DISE) in children was introduced as a method to evaluate the UA of children with complex UA obstruction or persistent OSA after AT [8,13,14]. It is a safe and cost-effective tool to evaluate the level and degree of UA obstruction during drug-induced sleep [15,16]. In contrast to cine-MRI, DISE allows direct visualization of the airway at multiple levels during spontaneous breathing, demonstrating the type and degree of airway obstruction in a clinical setting, and the findings may direct further surgery without the need of another sedation or admission to the hospital [8]. Previous reports in children with complex UA obstruction describe a heterogeneous population including children with DS, but also children with other medical conditions predisposing to UA collapse and children with or without a history of previous UA surgery [9,15,17].

This study aims, first, to describe the pattern of UA obstruction during DISE in a group of children with DS and OSA, without previous UA surgery; and, second, to evaluate the outcome of DISE-directed treatment and to investigate which DISE findings are related to failure of UA surgery.

2. Methods

A prospective study was performed including all children with DS who are enrolled in our multidisciplinary Down team (Antwerp University Hospital, Belgium), seen at the Ear Nose Throat (ENT) department and diagnosed with OSA. This study was approved by the local Ethics Committee (B30020108389). All parents gave written informed consent. Children with previous UA surgery [(adeno)tonsillectomy] were excluded from the study.

In each child, a clinical examination was performed, and tonsil size was scored according to the Brodsky score [18]. Body mass index (BMI) *z* scores were calculated according to Flanders growth curves for boys and girls.

Diagnosis of OSA was based upon full-overnight polysomnography (PSG), performed at the Pediatric Sleep Disorders Center of the Antwerp University Hospital, Belgium. The following variables were continuously measured and recorded by a computerized polysomnography device (Brain RT, OSG, Rumst, Belgium): electroencephalography (C4/Al, C3/A2, F3/A2, F4/A1), 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. If snoring was present during the night, this was reported (as present or absent). Polysomnograms were manually scored by certified technicians according to international guidelines [19].

A diagnosis of OSA is established with an obstructive apnea–hypopnea index (oAHI) ≥ 2/h [20]. Obstructive sleep apnea severity was defined as mild with oAHI ≥2/h and <5/h, moderate with oAHI ≥5/h and <10/h, and severe OSA with an oAHI ≥10/h.

Drug-induced sedation endoscopy was performed in the operating theater by a single pediatric ear–nose–throat surgeon, as previously described [21]. The children were sedated on the operating table with full cardiovascular monitoring by a pediatric anesthesiologist, using facial mask with a mixture of sevoflurane and oxygen. When intravenous access was obtained, sevoflurane was stopped and intravenous propofol was administered with a bolus injection of 1–2 mg, followed by continuous infusion according to body weight (6–10 mg/kg/h) to obtain the desired level of sedation and to maintain spontaneous breathing. Once a stable breathing pattern was obtained, a flexible fiberoptic laryngoscope was passed through a swivel adaptor on the mask and introduced into one nostril up to the level of the nasopharynx. No local anesthesia was used, and care was taken to avoid any pressure from the mask on the patient's face. The examination was performed with the child lying supine and the head in a neutral position. From the nasopharynx, the scope was gently passed toward the oral cavity, hypopharynx, and larynx. At each level, the pattern of UA collapse was scored according to a standard protocol [21]. The presence and degree of fixed and dynamic airway obstruction were noted.

At each UA level, the degree of obstruction was described. At the level of the nasopharynx, adenoid hypertrophy was graded as follows: 0, no adenoids; 1, adenoids occupying <50% of the lumen; 2, adenoids occupying 50–75% of the lumen; and 3, adenoids >75% obstruction of the nasopharynx by adenoid tissue. A score ≥2 was considered significant adenoid hypertrophy. Tonsillar obstruction was graded as 0 when there were no tonsils present; 1, with <50% collapse of the tonsils; 2, with 50–90% collapse of the tonsils; and 3, with tonsils touching at the midline. A score ≥2 was considered as indicating significant tonsillar hypertrophy. Tongue base obstruction was always in anteroposterior direction and scored as absent (0), partial collapse (1), or complete collapse (2).

Dynamic obstructions include hypotonia, defined as follows: a circumferential collapse at the entire oropharyngeal and/or hypopharyngeal level; anteroposterior palatal collapse or flutter; an anteroposterior collapse of the epiglottis, which is sucked against the posterior pharyngeal wall; and findings of late-onset laryngomalacia. The latter is characterized by redundant mucosa of the aryepiglottic folds being pulled into the airway and causing UA obstruction during forceful inspiration (type 1 laryngomalacia). Dynamic collapse was either absent (0) or present (1) for the following parameters: anteroposterior palatal collapse, collapse of the epiglottis, laryngomalacia, and hypotonia at oropharyngeal of hypopharyngeal level [21].

Multi-level obstruction was defined as the presence of one or more UA abnormalities outside the adenotonsillar region. When DISE showed clinically relevant obstruction at the level of the adenoids or tonsils, the child was intubated and ventilated at the end of the endoscopic evaluation, and UA airway surgery, ie, (adeno)/tonsillectomy, was performed during the same anesthesia. Surgery was performed with cold instruments; in case of tonsillectomy, the anterior and posterior tonsillar pillars were sutured with resorbable sutures. If no surgical intervention was required, the child was awakened following the DISE examination and transferred to the recovery room.

All children undergoing surgery were scheduled for a control PSG at least three months postoperatively. Surgical intervention was considered successful with a postoperative oAHI <5/h, and a complete cure was defined with a postoperative oAHI <2/h. An overview of the decision-making process is depicted in Fig. 1.

In the case of persistent disease (oAHI ≥5/h), additional treatment was based on OSA severity and preoperative DISE findings. A third PSG was scheduled to evaluate additional treatment outcome.

Statistical analysis was performed using IBM SPSS statistical version 20. Data are reported as median value with lower and upper quartiles unless otherwise stated. Correlations between variables were calculated using the Spearman correlation coefficient, and pre- and postoperative data were compared by the Wilcoxon signed-rank test. Statistical significance was obtained at a p value of <0.05.

3. Results

A total of 41 children with DS who met the inclusion criteria underwent DISE between September 2010 and June 2015. The study group consisted of 18 boys and 23 girls, aged 4.2 years (range,