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Original article

Contribution of drug-induced sleep endoscopy to the management of pediatric obstructive sleep apnea/hypopnea syndrome

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ABSTRACT

Objectives: The role of drug-induced sleep endoscopy (DISE) in the management of obstructive sleep apnea/hypopnea syndrome (OSAHS) is not precisely defined in children. The primary objective of this study was to describe DISE-induced revision of airway obstruction site location and the ensuing treatment changes in children with OSAHS. Secondary objectives were to analyze the correlation of number of obstruction sites found on DISE with apnea-hypopnea index (AHI) and with type of OSAHS.
Material and methods: A retrospective single-center study included 31 children (mean age: 5.5 ± 2.6 years) undergoing DISE for management of OSAHS between 2015 and 2018. Revisions of airway obstruction site location and in treatment were noted. The correlation of number of obstruction sites with AHI and with type of OSAHS was analyzed.
Results: Airway obstruction site location was reconsidered in 77% of children ($n = 24$), modifying treatment in 45.2% ($n = 14$). There was no significant correlation between number of obstruction sites and AHI: Spearman coefficient 0.20 ($P = 0.26$). Patients with type-III OSAHS did not show more obstruction sites than others: respectively, 2.0 versus 1.8 ($P = 0.40$).
Conclusion: DISE induced significant revision of the location and change in treatment of obstruction sites in children with OSAHS. Systematic implementation, especially in type-I OSAHS, would allow more precise pre-therapeutic classification and treatment adapted to actual airway obstruction.

1. Introduction

Prevalence of obstructive sleep apnea/hypopnea syndrome (OSAHS) in children is 1.2-5.7% [1,2], peaking between 2 and 6 years of age. Diagnosis is clinical and paraclinical, based on interview, questionnaires, physical examination and sleep recording [3]. The reference paraclinical examination is overnight polysomnography (PSG) in a sleep laboratory [4], which can be replaced by simplified recordings such as respiratory polygraphy (RP) conducted under precisely defined conditions [3]. These examinations can identify and quantify OSAHS, but do not provide information on the pharyngeal or laryngeal obstruction site or sites. Palatine tonsil and adenoid hypertrophy is usually implicated, and the most frequent treatment is adenotonsillectomy (AT). However, residual postoperative OSAHS, found in 30–70% of cases [5–7] testifies to the limitations of preoperative obstruction assessment. Drug-induced sleep endoscopy (DISE) consists in flexible endoscopic exploration of the pharynx and glottic and supraglottic larynx under anesthesia mimicking physiological sleep. The aim is to identify any occult obstruction sites overlooked by awake clinical examination [8]. Indications for DISE in pediatric sleep disordered breathing (SDB) remain poorly defined. It seems to be useful when awake clinical examination is non-contributive [9], in residual OSAHS after AT, and in case of craniofacial deformity or neuromuscular involvement (type-III OSAHS on the Capdevila-Gozal classification [10]), in which multiple obstruction sites are more frequent.

The main objective of the present study was to assess revision of obstruction site location induced by DISE and the consequent change in treatment of children with OSAHS. The first secondary objective was to assess correlation between the number of sites found on DISE and preoperative apnea-hypopnea index (AHI); the second was to compare site number between type-III and other OSAHS types.

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2. Material and methods

2.1. Population

A single-center retrospective study included 70 consecutive children undergoing DISE in the ENT and Head and Neck Surgery Department of the University Hospital of Montpellier, France, between October 2015 and June 2018.

Inclusion criteria comprised DISE performed for OSAHS confirmed on sleep recording; and age < 12 years at DISE. Nine children were excluded for age > 12 years, 16 for missing sleep recording data (examination not performed before DISE or data missing in examination report), 13 for OSAHS not confirmed on sleep recording, and 1 for missing clinical data (patient managed elsewhere). Analysis thus included 31 files.

Mean age was 5.5 ± 2.6 years (range, 10 months to 11 years), with 19 boys. Eighteen patients (58%) presented with type-III OSAHS: 5 with hypotonia, 8 with craniofacial deformity, and 5 with Down syndrome. The others had no particular history, notably of overweight (type-II OSAHS) and were therefore counted as type-I (i.e., not type-II or III) on the Capdevila-Gozal classification [10], despite no clear adenotonsillar lymphoid tissue hypertrophy.

All patients were examined by a senior pediatric ENT physician ahead of DISE. On interview, 74% of parents (n = 23) reported snoring, 39% (n = 12) nocturnal apnea, and 55% (n = 17) indirect signs of sleep apnea: night sweats, daytime asthenia or agitation, enuresis despite acquisition of nocturnal continence, or morning headache. Potentially involved obstruction sites were noted: 71% of children (n = 22) had tonsillar volume equal to or less than Brodsky grade 2 [11], and 29% (n = 9) had grade 3 or 4 hypertrophy. In 74% of cases (n = 23), physical examination was completed by nasal flexible endoscopy (NFE), detecting adenoid obstruction in 12 cases, suspected tongue-base obstruction in 2 (patients 1 and 16), and suspected occult laryngomalacia in 1 (patient 12).

DISE was indicated in 58% of cases (n = 18) as part of the preoperative work-up in type-III OSAHS (NFE previously performed in 13 cases), in 26% (n = 8) for residual OSAHS after tonsillectomy and/or adenoidectomy (NFE previously performed in 5 cases), and in 16% (n = 5) for OSAHS confirmed on sleep recording without obstruction site found on awake clinical examination(NFE previously performed in all 5 cases).

2.2. Sleep recording

Sleep recording was systematic ahead of DISE, with either overnight polysomnography in a sleep laboratory (26%, n = 8), or respiratory polygraphy, in the ENT department for over-2 year-olds (58%, n = 18), or in the pediatrics department for under-2 year-olds (16%, n = 5). Interpretation followed the American Academy of Sleep Medicine (AASM) 2012 criteria [12,13]. Study data comprised: apnea/hypopnea index (AHI), obstructive apnea/hypopnea index (OAHI), and oxygen desaturation index (ODI). There is at present no consensus on normal values in childhood OSAHS; we considered OAHI > 1.5/h as pathological [14].

Median AHI was 4.8 (interquartile range (IQR): 3.3–7.5), median OAHI 3.8 (2.7–6.3), and median ODI 7.1 (3.6–15.3). All patients had OAHI > 1.5.

2.3. DISE

DISE was performed in theater by the senior pediatric ENT surgeon who had performed the preoperative clinical assessment. The child was positioned supine, in a quiet room in the shade. Sleep was induced by pediatric anesthetists under cardiorespiratory monitoring.

Anesthesia protocols were systematically noted (Table 1): inhaled induction anesthetic (sevoflurane) ahead of venous line, intravenous (IV) anesthetics to maintain induced sleep. Forty two

Table 1
Anesthesia protocols used in DISE.

Patients	Induction by mask: Sevoflurane	Induced sleep maintenance	Molecule
1	X	IV	Propofol/Remifentanil hydrochloride
2	X	Inhalation	Sevoflurane
3	X	Inhalation	Sevoflurane
4	X	IV	Propofol/Remifentanil hydrochloride
5	X	IV	Propofol
6	X	IV	Propofol/Remifentanil hydrochloride
7	X	IV	Propofol/Remifentanil hydrochloride
8	X	IV	Propofol
9		IV	Propofol/Sufentanil
10		IV	Propofol/Remifentanil hydrochloride
11		IV	Propofol/Remifentanil hydrochloride
12		IV	Propofol/Remifentanil hydrochloride
13		IV	Propofol/Remifentanil hydrochloride
14		IV	Propofol/Remifentanil hydrochloride
15		IV	Propofol/Remifentanil hydrochloride
16		IV	Propofol
17	X	IV	Propofol
18		IV	Propofol/Remifentanil hydrochloride
19	X	IV	Propofol/Remifentanil hydrochloride
20		IV	Propofol/Remifentanil hydrochloride
21		IV	Propofol/Remifentanil hydrochloride
22		IV	Propofol/Remifentanil hydrochloride
23	X	IV	Propofol/Remifentanil hydrochloride
24	X	IV	Propofol/Remifentanil hydrochloride
25	X	IV	Propofol/Remifentanil hydrochloride
26		IV	Propofol/Remifentanil hydrochloride
27		IV	Propofol/Remifentanil hydrochloride
28		IV	Propofol/Remifentanil hydrochloride
29		IV	Propofol/Remifentanil hydrochloride
30	X	IV	Propofol/Remifentanil hydrochloride
31		IV	Propofol

IV: intravenous.

percent of patients ($n = 13$) had sevoflurane mask induction. Ninety three percent ($n = 29$) had IV maintenance. IV molecules comprised propofol associated to remifentanil hydrochloride in 74% of cases ($n = 23$), propofol alone in 17% ($n = 5$), and propofol associated to sufentanil in 3% ($n = 1$). Two patients (6%) had induction and maintenance by sevoflurane inhalation alone.

The pediatric flexible endoscope (2.8 × 270 mm, Karl Storz & Co, Tuttlingen, Germany) was introduced in one nostril without local anesthesia. Obstruction was assessed in the nasal cavity, pharynx and larynx. In case of abnormality in the supraglottic or glottic larynx during DISE, rigid laryngotracheal endoscopy was associated. Obstruction per site was assessed as non-obstructive, partially obstructive or totally obstructive. In case of velar, tongue-base or epiglottic obstruction, mandibular advancement was performed.

The VOTE classification [15] was used, plus description of nasopharynx and larynx. Obstruction was considered multi-site when more than 1 site was found on DISE, whether obstruction was total or partial.

Revision of obstruction location was defined by:

- discovery of a site not found on awake physical examination; adenoid hypertrophy discovered on DISE in patients who had not had prior awake NFE was not considered as a DISE-induced change;
- and/or confirmation of a suspected site, as suspicion on awake examination was not in itself enough to indicate adapted surgical treatment;
- and/or elimination of a suspected site on awake examination.

Location revision could be followed either by surveillance or by treatment modification: surgery at the new site, confirmation or cancellation of initially planned surgery, or implementation of continuous positive airway pressure (CPAP).

2.4. Statistical analysis

Correlation between number of sites on DISE and AHI was assessed on Spearman correlation coefficient. Difference in AHI according to single- or multi-site involvement was assessed on Wilcoxon signed ranks test. Difference in number of sites according to type-III and "type-I (by default)" OSAHS was assessed on Wilcoxon signed ranks test.

3. Results

3.1. Change in treatment following DISE

No complications occurred, no intubation was required, and DISE could be performed as planned in all cases. Snoring was found in 97% of patients ($n = 30$).

Only 1 DISE examination did not find snoring, and NFE had found no obstruction site in this particular patient, who was 9 years old at examination, with OSAHS confirmed on PSG (AHI 6.9, ODI 11.8) performed as part of exploration of restless legs syndrome associated with attention deficit/hyperactivity disorder; interview had revealed no signs of nocturnal obstruction or indirect signs of OSAHS; physical examination had found no obvious obstruction site; the patient had history of palatine tonsillectomy 2 years previously. The anesthesia protocol comprised induction and parenteral maintenance by propofol and remifentanil hydrochloride. Control PSG was scheduled for 1 year post-DISE, as sleep quality had improved with treatment for the restless legs syndrome.

DISE was completed by pre-scheduled palatine tonsillectomy and/or adenoidectomy in 21 children: 7 isolated adenoidectomies (22%), 3 isolated palatine tonsillectomies (10%) and 11 combined procedures (35%).

DISE led to revision of obstruction location in 77% of cases ($n = 24$). Table 2 shows clinical and paraclinical data and post-DISE management. Six of the patients had not had initial NFE, and 4 of these showed adenoid hypertrophy on DISE. DISE identified new sites in 15 children, resulting in surveillance in 11 cases (4 velar, 2 epiglottic and 7 tongue-base sites) and change in treatment in 4: 2 tonsillectomies, 1 aryepiglottic fold division (patient 8), and 1 epiglottopexy (patient 17). In 4 cases, a suspected site was confirmed, and treated in all cases: 1 aryepiglottic fold division (patient 12), 3 palatine tonsillectomies. In 1 case (patient 2), a tonsillar site was ruled out, and the planned palatine tonsillectomy was cancelled. In 1 case (patient 29), a new site (tongue base) was identified and put under surveillance and a suspected site (palatine tonsils) was confirmed and operated on. In 1 case (patient 1), a tongue-base site was ruled out and a suspected tonsillar site was confirmed and operated on (palatine tonsillectomy). In 2 cases (patients 7 and 26) 1 or more new sites were identified (velar palate, tongue base, epiglottis) and a tonsillar site was ruled out, with cancellation of palatine tonsillectomy. Excluding new sites put under simple surveillance, DISE altered treatment in 14 cases (45.2% of the series).

In patients 1 and 16, the tongue-base obstruction suspected on nasal endoscopy was respectively ruled out and confirmed. In patient 12, the occult laryngomalacia suspected on nasal endoscopy was confirmed in DISE and treated, by aryepiglottic fold division.

Considering each group of DISE indication, obstacle sites were revised in 83% of preoperative assessments for type-III OSAHS ($n = 15/18$), 62% of cases of persistent OSAHS after adenotonsillectomy ($n = 5/8$) and 80% of patients without obstruction site on awake examination ($n = 4/5$).

In 7 cases (23%), DISE led to no site revision or change in treatment.

3.2. Correlation between site number on DISE and AHI

71% of patients ($n = 22$) showed multi-site obstruction, with median AHI of 4.2 (IQR, 3–7.7), compared to 5.7 (4.3-7.8) in case of single-site obstruction: i.e., no significant difference in AHI between the two groups; ($P = 0.45$).

There was no significant correlation between site number on DISE and preoperative AHI: Spearman correlation coefficient, 0.20 ($P = 0.26$).

3.3. Site number according to type-III or default type-I OSAHS

Fig. 1 shows obstruction location according to OSAHS type. In type III ($n = 18$), 4 children showed epiglottic obstruction by posterior tilt and 6 showed tongue-base obstruction (Fig. 2). In the 13 children classified as type I by default, there was a single epiglottic obstruction and 4 tongue-base 4 obstructions.

Mean site number was non-significantly greater in type III than "type I by default": 2.0 compared to 1.8 ($P = 0.40$). DISE impact on site location was likewise comparable: 83% revision ($n = 15/18$) compared to 62% ($n = 8/13$), respectively.

In case of multiple obstruction sites, the most severe obstacle was first removed (except for palatine tonsils and adenoids, treated in 1 step), with clinical and paraclinical screening for any residual OSAHS. Excluding the 5 patients without obvious obstruction site(and thus without initial surgical target) on awake examination, DISE revised the main obstructive site location in 8 out of 26 cases (30.1%).

4. Discussion

Our patients were divided in 3 groups according to indication for DISE.

Table 2
DISE-induced revision of airway obstacle locations.

Clinical and paraclinical data							DISE			
Patients	Gender	Age at DISE (months)	Context/indication for DISE	AHI	Awake endoscopy	Obstacle(s) on awake examination	Obstacle(s) on DISE	Type of DISE-induced revision	Treatment	Revision of main obstacle
1	M	73	OSAHS type III (craniofacial deformity)	4.4	Yes	Palatine tonsil (P)-Tongue base (P)	Palatine tonsil (C)	2-3	Tonsillectomy	No
2	M	76	Residual OSAHS after surgery	4.1	No	Palatine tonsil (P)	Adenoids (C)	3	Adenoidectomy-Cancellation Tonsillectomy	Yes
3	M	33	No obvious obstruction site	1.9	Yes	Adenoids (P)	Adenoids (P) - Palatine tonsil (C)	1	AT	Yes
5	M	88	No obvious obstruction site	2.8	Yes	Adenoids (P)	Adenoids (P)-Epiglottis (P)	1	Adenoidectomy-Surveillance epiglottis	No
6	F	51	OSAHS type III (craniofacial deformity)	4.2	Yes	Adenoids (C) - Palatine tonsil (C)	Adenoids (C) - Palatine tonsil (C) - Tongue base (P)	1	AT- Surveillance tongue base	No
7	F	88	OSAHS type III (craniofacial deformity)	20	Yes	Palatine tonsil (P)	Velar palate (C) - Epiglottis (C)	1-3	Nocturnal CPAP - Cancellation Tonsillectomy-Surveillance velar palate and epiglottis	Yes
8	F	65	No obvious obstruction site	3.7	Yes	None	Velar palate (P) -Laryngomalacia (C)	1	AEF division-Surveillance velar palate	No
9	F	86	Residual OSAHS after surgery	2.9	No	Palatine tonsil (C)	Adenoids (P) - Velar palate (P) - Palatine tonsil (C)	1	AT-Surveillance velar palate	No
11	M	138	OSAHS type III (T21)	20.7	Yes	Adenoids (C) - Palatine tonsil (P)	Adenoids (C) - Palatine tonsil (C)	2	AT	Yes
12	M	10	No obvious obstruction site	2.7	Yes	Suspected occult laryngomalacia	Laryngomalacia (C)	2	AEF division	No
13	F	90	Residual OSAHS after surgery	5.7	Yes	None	Velar palate (P) - Tongue base (C)	1	Surveillance velar palate and tongue base	No
14	F	45	OSAHS type III (craniofacial deformity)	7.7	Yes	Adenoids (C)	Adenoids (P) - Palatine tonsil (C)	1	AT	Yes
16	M	110	Residual OSAHS after surgery	2.3	Yes	Tongue base (P)	Velar palate (C) - Tongue base (P)	1	Surveillance velar palate and tongue base	No
17	F	19	OSAHS type III (Hypotonia)	7	Yes	None	Epiglottis (C)	1	Epiglottopexy	No
18	F	29	OSAHS type III (T21)	1.8	Yes	Adenoids (P) - Palatine tonsil (C)	Adenoids (P) - Palatine tonsil (C) - Tongue base (P)	1	AT-Surveillance tongue base	No
19	M	50	OSAHS type III (Hypotonia)	6.4	No	Palatine tonsil (C)	Adenoids (P) - Palatine tonsil (C) - Epiglottis (P)	1	AT-surveillance epiglottis	No
21	M	54	Residual OSAHS after surgery	27	Yes	Palatine tonsil (C)	Palatine tonsil (C) - Tongue base (C)	1	Tonsillectomy-surveillance tongue base	No
22	M	27	OSAHS type III (craniofacial deformity)	12.7	No	None	Adenoids (P) - Palatine tonsil (C)	1	AT	No
24	M	59	OSAHS type III (Hypotonia)	12.1	Yes	Adenoids (C)	Adenoids (C) - Tongue base (C)	1	Adenoidectomy-Surveillance tongue base	No

Table 2 (*Continued*)

Clinical and paraclinical data							DISE			
Patients	Gender	Age at DISE (months)	Context/indication for DISE	AHI	Awake endoscopy	Obstacle(s) on awake examination	Obstacle(s) on DISE	Type of DISE-induced revision	Treatment	Revision of main obstacle
26	M	65	OSAHS type III (T21)	11.6	Yes	Adenoids (C) - Palatine tonsil (P)	Adenoids (C) - Tongue base (C)	1-3	Adenoidectomy-Cancellation Tonsillectomy-Surveillance tongue base	Yes
27	F	79	OSAHS type III (craniofacial deformity)	10	No	Palatine tonsil (P)	Palatine tonsil (C)	2	Tonsillectomy	No
28	M	62	OSAHS type III (craniofacial deformity)	3	No	Palatine tonsil (C)	Palatine tonsil (C) - Tongue base (C)	1	Tonsillectomy-Surveillance tongue base	No
29	M	61	OSAHS type III (T21)	5.3	Yes	Adenoids (P) - Palatine tonsil (P)	Adenoids (P) - Palatine tonsil (C) - Tongue base (P)	1-2	AT-Surveillance tongue base	Yes
30	M	102	OSAHS type III (Hypotonia)	4.1	Yes	Adenoids (P) - Palatine tonsil (P)	Adenoids (P) - Palatine tonsil (C)	2	AT	Yes

1: discovery of new obstruction site; 2: confirmation of site suspected on awake examination; 3: suspected on awake examination ruled out; (C): complete obstruction; (P): partial obstruction; AT: adenotonsillectomy; AEF: aryepiglottic folds.

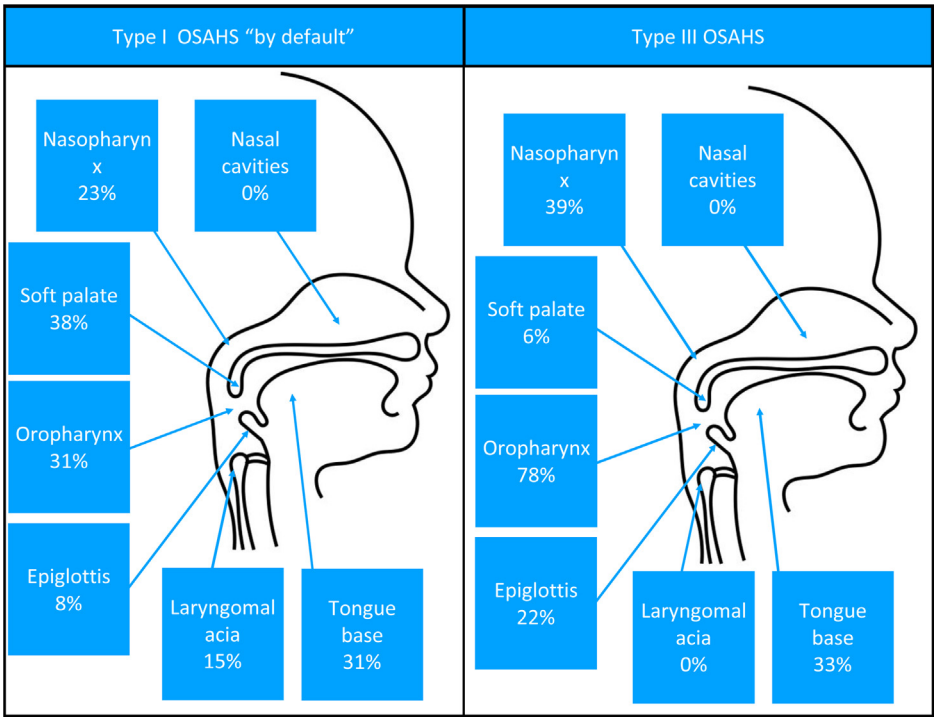

Fig. 1. Distribution of obstruction sites on DISE in "type I by default" and in type III OSAHS.

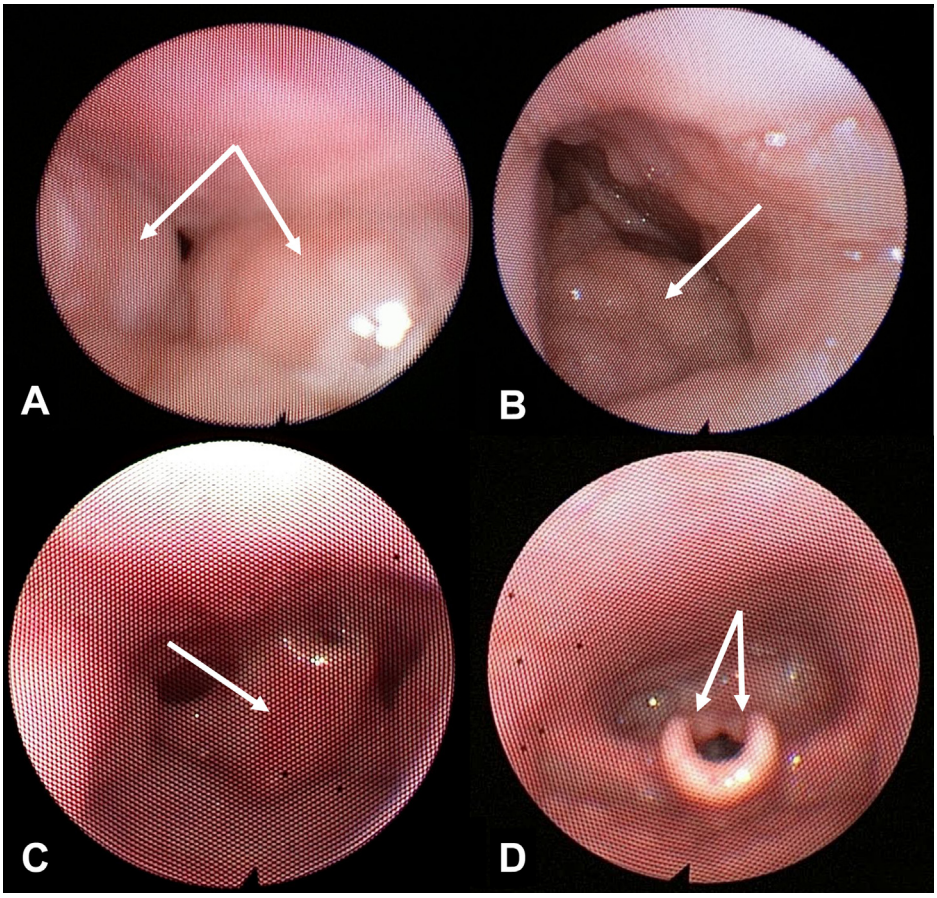

Fig. 2. Photographs of induced sleep endoscopies. A: transverse oropharyngeal obstruction related to palatine tonsils (white arrows) assessed as Brodsky grade 2 on awake examination. B: tongue-base obstruction with apparent lingual tonsil hypertrophy (white arrow) non-detected on awake examination. C: obstruction by posterior tilt of epiglottis (white arrow) without associated tongue-base retraction. D: supraglottic obstruction with short aryepiglottic folds and anterior tilt of arytenoids on inbreath (white arrows).

The first indication was to screen for an obstruction site after failure of adenotonsillectomy. In case of risk factors for failure of adenotonsillectomy in OSAHS (age > 7 years, asthma, or type II or III OSAHS [16]), certain authors [17] recommend systematic prior DISE.

The second indication was to confirm an obstruction site when physical examination was insufficiently contributive. Here, DISE can reveal velar, retro-basilingual or supraglottic obstructions that are often undetected on awake examination, and can confirm indications for aryepiglottic division (2 cases in the present series), epiglottopexy (1 case), lingual tonsillectomy, pharyngoplasty or orthodontics. In the particular case of Brodsky grade ≤ 2 tonsil size, it can confirm or rule out palatine tonsillar obstruction during sleep (Fig. 2). This choice of a grade 2 threshold indicating DISE in the presort study is open to debate, as there is no consensus in the literature. Moreover, assessment of tonsillar obstruction in DISE and resulting surgical indications are subjective and qualitative. If tonsillectomy is cancelled following DISE, it is important to institute systematic follow-up with reassessment of obstruction signs [3]. Parents should also be clearly informed of potential progression of symptoms and tonsil volume and the possibility of later tonsillectomy founded on stronger arguments. Lingual tonsillectomy is the second most frequent procedure, after adenotonsillectomy, in surgical treatment of childhood OSAHS [18]. It seems less effective in case of glossoptosis with craniofacial deformity [19] and of obesity [20], with symptom resolution rates ranging between 57% and 88% [19–23]. Occult laryngomalacia manifests after 2 years of age in the form of snoring, interview suggesting apnea without stridor, feeding problems or failure to thrive [24]. Prevalence in childhood OSAHS is 3.9%, with frequent association with other obstruction sites [25]; in the present series, prevalence was comparable, with 2 cases (Fig. 2). DISE is strongly contributive to detection. Chan et al. [26] reported that supraglottoplasty improved AHI in 91% of cases of occult laryngomalacia.

The third indication is to screen for multi-site involvement, which is more frequent in type-III OSAHS [27], although this was not confirmed in the present series, probably due to sample size and the pre-DISE clinical characteristics of the other children; even so, type-III OSAHS requires precise assessment of potential sites. As well as craniofacial deformity and impaired tonus, these children often have severe comorbidity, requiring certainty in diagnosis before surgery can be considered: hemorrhagic risk, vascular malpositioning, and postoperative pain and feeding management. DISE enables surgery to target only obstruction confirmed on sleep endoscopy and actually implicated in the OSAHS. In certain pathologies such as Down syndrome, in view of the high rate of residual OSAHS following adenotonsillectomy, DISE tends to be systematic in the surgical decision tree [28].

DISE found no obstruction site in only 1 case; either the anesthesia protocol failed to recreate the physiological sleep conditions in which apnea occurs, or the PSG had been performed during an intercurrent nasopharyngeal episode exacerbating the child's respiratory status.

In the literature, obstruction sites found on awake endoscopy and DISE correlate poorly [8,29]. Oropharyngeal, basilingual and supraglottic sites are underestimated on awake endoscopy, which does not take account of modified tonus during sleep, so that assessment in consultation does not correspond to the degree of obstruction experienced during sleep. Awake endoscopy does, however, reliably assess nasal obstruction and adenoid volume [29]. We therefore did not count discovery of adenoid obstruction as revision on DISE in the 4 patients who had not undergone prior awake nasal endoscopy, as it would doubtless have detected this hypertrophy if it had been performed.

Anesthesia-induced hypotonia during DISE may lead to overestimation of obstruction sites, raising the question of the optimal anesthesia protocol. In the present series, protocols varied. Propofol is currently most widely used, due to its short half-life and the absence of side-effects of nausea and vomiting [30]. Recent studies showed that ketamine and dexmedetomidine induced less oxygen desaturation, enabling better maintenance of blood pressure and better conditions for DISE than propofol or propofol associated to sevoflurane [31]. Dexmedetomidine, however, is an expensive molecule that is subject to special authorization in France for use in the operating theater in children.

Our choice to use RP rather than the standard PSG in 18 cases is open to question; it was determined by considerations of logistics and wait time [3], especially relevant in pediatrics. Even so, the French Health Authority (HAS) considers that RP can replace PSG in the exploration of childhood OSAHS when performed and interpreted by a specialized team [4]. Like Boudewyns et al. [31], we found no correlation between AHI elevation and multi-site involvement; in contrast, Ulualp et al. [32] reported that moderate to severe OSAHS was usually associated with multi-site involvement. In case of multiple sites, we treated the most obstructive first. Indeed, given airway flow dynamics, it is not impossible that detection of "secondary" sites on DISE was increased by the principal obstruction site.

By analogy with DISE in adults, we used the VOTE classification [15], adding nasal obstruction, adenoid volume and laryngomalacia screening. There is no consensus as to classifications [27]. The classification recommended by Chan et al. seems adapted to pediatric populations [33]; the authors identify 5 sites (adenoid volume, velar palate, palatine tonsil, lingual tonsil, and supraglottic level), with 4 degrees of obstruction for each. At present, there are at least 6 scoring systems in use [34]. Fishman et al. [30] use their own classification, not taking velar obstruction into account, but including a potential nasal cavity site. In the present series, there were no nasal obstructions, probably as the two cavities were not both checked systematically at the beginning of DISE. It does, however, seem to be an interesting site to assess, notably in view of the prevalence of inferior turbinate hypertrophy found in a recent cohort of 150 children treated for OSAHS [35]. International standardization of DISE scoring in children would help harmonize practices and facilitate comparison across studies.

In 2008, Capdevila and Gozal reported that children with OSAHS and history of obesity showed extra risk of comorbidity, a particular clinical presentation and increased risk of persistent OSAHS after adenotonsillectomy [10]. They thus distinguished 2 OSAHS types: children with normal weight and obstructive syndrome related to lymphoid tissue hypertrophy (type I OSAHS), or children with obesity (type II). A third type was defined by craniofacial or muscular abnormality, again associated with risk of persistent OSAHS (type III). Management of moderate to severe persistent OSAHS at that time included overnight CPAP; subsequently, the introduction of pediatric DISE enhanced exploration of the causes of OSAHS, revealing velar and basilingual obstructions, posterior epiglottic tilt and occult laryngomalacia, with specific treatments. The present "type I by default" group (i.e., not type II or III) shows that the Capdevila-Gozal classification can be improved upon, as this is not simple lymphoid tissue hypertrophy. In the light of recent literature findings, it seems advisable to include DISE systematically in the preoperative work-up for childhood OSAHS, modifying the classification, by keeping group II (obesity) and III (craniofacial or neuromuscular deformity), but dividing group I (no particular history, no obesity, no craniofacial or neuromuscular deformity) into 2 subgroups:

- group Ia: isolated clinical adenotonsillar lymphoid tissue hypertrophy underlying the DISE obstruction (i.e., group I as described in 2008);

- group Ib: other cause of obstruction with or without adenotonsillar lymphoid tissue hypertrophy on DISE (i.e., in the present report, "type I by default").

This is a pre-treatment classification whereby the interview already identifies type II or III OSAHS and clinical and complementary examination (PSG or RP as indicated, plus DISE) identifies type Ia or Ib. The advantage lies in being able to offer personalized treatment according to the obstructions detected on DISE, and thus reducing residual postoperative OSAHS. In type Ia, DISE could be performed under the same general anesthesia step as scheduled adenotonsillectomy: DISE followed by intubation, with surgery if palatine tonsil involvement is confirmed. In type Ib, DISE would be systematically included in the pre-treatment work-up. Any surgery (palatine tonsillectomy with Brodsky grade ≤ 2 volume, or surgery for occult laryngomalacia, epiglottic posterior tilt or basilingual obstruction) could be postponed to a later general anesthesia after informing parents and possibly performing complementary imaging.

5. Conclusion

DISE is a diagnostic complement to clinical examination, questionnaires and sleep recording in the management of childhood OSAHS. It is especially useful when clinical examination does not suggest adenoid or tonsillar involvement, in case of residual OSAHS after adenotonsillectomy, and in type-III OSAHS, where it revises airway obstacle locations in more than two-thirds of cases. There is a need to improve codified scoring of obstruction sites and to standardize anesthesia protocols. The clinical and paraclinical benefit of integrating DISE in the OSAHS decision-tree remains to be assessed in larger-scale prospective randomized studies.

Disclosure of interest

The authors declare that they have no competing interest.

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