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Drug-Induced sleep endoscopy in children: NAVOTEL scoring system development

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Abstract

Objectives: Pediatric drug-induced sleep endoscopy (DISE) lacks a universal and easy-to-use scoring system. The velum, oropharynx, tongue, epiglottis (VOTE) scoring system is widely used but needs to be completed in pediatrics. The main objective of this study was to investigate the distribution of obstructive sites in DISE and to propose an appropriate pediatric scoring system. The secondary objective was to evaluate the changes in surgical management induced by the proposed scoring system.

Methods: A single-center prospective 5-year study was conducted from March 2016 to December 2021, including 99 children with a mean age of 7.2 years (±3.7), with pathological preoperative sleep recordings and undergoing DISE. The distribution of all upper airway obstructive sites was studied.

Results: Adenoids (A) were the most frequent obstructive site (63% of patients), and the nasal cavities (N) and the larynx (L) were other frequent obstructive sites. These sites are not explored by the VOTE scoring system, leading to the creation of the nose, adenoids, velum, oropharynx, tongue, epiglottis, larynx (NAVOTEL) scoring system. NAVOTEL was significantly correlated with the severity of obstructive sleep apnea-hypopnea syndrome (OSAS) ($\rho = 0.2$; $p = 0.04$) and highlighted obstructive sites in 6/9 patients with VOTE = 0. Of these patients, 4 had a complete obstructive site, and 3 had a multisite obstruction. VOTE indicated 8 additional surgical actions; NAVOTEL indicated 50 other actions compared to clinical examination. The NAVOTEL scoring system was exhaustive regarding surgical indications for OSAS.

Conclusions: The NAVOTEL scoring system is exhaustive in pediatric DISE and correlated to OSAS severity. It should be preferred in pediatric DISE.

KEYWORDS

drug-induced sleep endoscopy, pediatric obstructive sleep apnea-hypopnea syndrome, pediatric sleep disordered breathing, tonsillectomy, VOTE classification

1 | INTRODUCTION

Pediatric obstructive sleep apnea-hypopnea syndrome (OSAS) has an estimated prevalence of 1%–4%.[1–3] Affected children may suffer daytime sleepiness, school difficulties, behavioral and neurocognitive problems, enuresis, cardiovascular complications, and metabolic and growth disorders.[4–8] OSAS in children significantly reduces the overall quality of life.[9]

Adenotonsillar hypertrophy is the most critical contributor to OSAS in children with no medical history; tonsillectomy with adenoidectomy is the first-line treatment, resulting in the resolution of symptoms in most affected children.[3,10,11] For treatment failures or children with a more complex history, a more sophisticated diagnostic approach is required for individualized management.

Drug-induced sleep endoscopy (DISE), first described in adults by Croft and Pringle[12] in 1991, is an examination performed under drug-induced sleep that enables the targeting of obstructive sites hidden on clinical examination and found to be obstructive during sleep. DISE in pediatrics has some specificities. However, most practitioners record their results using the velum, oropharynx, tongue, epiglottis (VOTE) scoring system created for adults.[13,14] In 2021, the American Academy of Otolaryngology-Head and Neck Surgery[15] highlighted the importance of documenting the anatomical sites and severity of obstruction, agreed that the nasal cavity, nasopharynx, palate, and soft palate, pharyngeal airway (including lateral oropharyngeal wall and base of tongue), and supraglottic larynx should be examined and concluded that there is a need to develop a pediatric DISE scoring system that can be universally adopted and used to enable better data collection, standardization of investigations, development of diagnostic algorithms, and ultimately improve therapeutics.

The main objective was to study the distribution of the obstructive sites found during DISE in children and to develop an adapted scoring system. The secondary objective was to analyze the changes in therapeutic management resulting from this new scoring system.

2 | METHODS

This is a prospective monocentric study, including all patients under 18 years of age with pathological sleep recording who underwent DISE between March 2016 and December 2021, that is, 69 months (5 years and 9 months), within a pediatric ENT and cervicofacial surgery department of a tertiary-care center. The institutional review board approved the study design (#2019-115). Informed consent was obtained from the parents or the legal representative. Pathologic sleep recording was defined by an obstructive apnea-hypopnea index (OAHI) of more than 1.5 obstructive apnea-hypopnea per hour (OAH/h), following the French Society of Otolaryngology-Head & Neck Surgery recommendations.[16] The exclusion criterion was the failure to perform DISE.

All children were examined by a senior pediatric ENT physician, who looked for obstructive sites in clinics after questioning about signs of OSAS. Tonsillar hypertrophy was assessed according to the Brodsky classification.[17] Fiberoptic laryngoscopy was performed when required. OSAS was classified into three types according to Capdevilla and Gozal[4]: type I—isolated lymphoid tissue hypertrophy; type II—obese patient (BMI > 95th percentile); type III—patient with a craniofacial malformation or a neuromuscular disorder.

Sleep recording consisted of polysomnography in a Sleep Department or respiratory polygraphy at home. Interpretations were made by AASM (American Academy of Sleep Medicine) standards.[18,19]

2.1 | DISE modalities

The indications for DISE were the search for the obstructive site in the face of an inconclusive clinical examination, a check-up after unsuccessful surgical treatment of OSAS, or a systematic assessment of obstructive sites in type III OSAS.

The senior ENT specialist who examined the child during the preoperative consultation performed the DISE in the operating room. The child was placed in the supine position. Pediatric anesthetists induced sleep under cardio-neuro-respiratory monitoring. The anesthetic protocol was identical for all patients, consisting of a naso-buccal mask induction of sevoflurane followed by intravenous maintenance with propofol. Once the child was well asleep, the fiberoptic nasolaryngoscopic examination was performed without local anesthesia.

All possible obstructive sites from the nasal cavity to the glottic plane were identified and scored in a standardized operative report as normal (0/2), partially obstructive (1/2), or completely obstructive (2/2). The sites included in the VOTE[13] scoring system were analyzed (Velum, Oropharynx, Tongue, and Epiglottis) and completed by pediatric-specific obstructive sites: the nasal cavity, the adenoids, and the larynx using the same scoring system. In the case of doubt of subglottic obstruction, the examination was completed by a rigid laryngoscopy.

2.2 | Statistical analysis

The Kruskal–Wallis test was performed for the distribution of the obstructive sites according to the OSAS type, the χ^2 test for the gender distribution, and the Pearson correlation test for the correlation of DISE scores with the OAHI. Bivariate analysis by linear regression was performed, with the outcome variable OAHI and the explanatory variables being the scoring systems, taking age as a confounding factor.

3 | RESULTS

One hundred and one patients underwent DISE after sleep recording during the study. Fifty-two males and 47 females were included, corresponding to a sex ratio of 1.1. Two patients were excluded for

TABLE 1 Characteristics of the series.

	Average (SD)	Median [Q25–75]	Min	Max	*n*
Age	7.23 (3.73)	6.70 [4.63; 9.70]	0.214	17.9	99
OAHI	6.55 (7.80)	4.50 [3.00; 7.00]	1.50	59.0	99
Number of sites	4.17 (1.07)	4.00 [4.00; 5.00]	0	7.00	99
Scores by site					
N	0.545 (0.674)	0 [0; 1.00]	0	2.00	99
A	0.960 (0.844)	1.00 [0; 2.00]	0	2.00	99
V	0.192 (0.509)	0 [0; 0]	0	2.00	99
O	0.899 (0.875)	1.00 [0; 2.00]	0	2.00	99
T	0.586 (0.769)	0 [0; 1.00]	0	2.00	99
E	0.343 (0.625)	0 [0; 1.00]	0	2.00	99
L	0.0808 (0.369)	0 [0; 0]	0	2.00	99
VOTE	2.02 (1.28)	2.00 [1.00; 2.00]	0	5.00	99
NAVOTEL	3.61 (1.66)	4.00 [2.00; 5.00]	0	8.00	99

Abbreviations: A, Adenoids; E, Epiglottis; L, Larynx; N, Nose; O, Oropharynx; OAHI, Obstructive Apnea-Hypopnea Index; V, Velum; T, Tongue.

failure to perform DISE: one for desaturation during the examination leading to oro-tracheal intubation and one for excess secretions preventing the anatomical analysis. 99/101 DISEs were retained for analysis.

3.1 | Patients

The characteristics of the patients are detailed in Table 1. Sixty-five percent ($n = 65$) of parents reported apnea episodes, 88% ($n = 88$) persistent snoring, and 96% ($n = 96$) indirect signs of OSAS. Awake fiberoptic nasolaryngoscopy was performed in 51% ($n = 51$) of the children, which revealed adenoidal hypertrophy in 55% ($n = 55$). Sixty-six percent of the children had type I OSAS, 14% had type II OSAS, and 20% had type III OSAS.

Sleep recording consisted of polysomnography in a Sleep Department for 27% of the patients ($n = 27$) or respiratory polygraphy at home for 73% ($n = 72$). The overall average OAHI was 6.6 OAH/h, with no significant differences between the groups.

3.2 | Obstructive site distribution

The most frequent obstructive site was the adenoids (63% of children), followed by the oropharynx (57%), and the nasal cavity (44%) (Table 2). The distribution of obstruction sites and scoring according to the OSAS type is shown in Table 3. A significantly different distribution according to OSAS type was noted for all obstructive sites except the nasal cavity and the larynx.

TABLE 2 Distribution of obstruction sites.

		N	A	V	O	T	E	L
Obstruction score	0	56%	37%	86%	43%	59%	74%	95%
	1	34%	29%	9%	23%	24%	18%	2%
	2	10%	33%	5%	33%	17%	8%	3%
% patients with obstruction	All	44%	63%	14%	57%	41%	26%	5%
	type I	45%	**57%**	**9%**	49%	**32%**	22%	5%
	type II	50%	79%	29%	57%	57%	29%	0%
	type III	40%	70%	20%	**80%**	60%	**40%**	10%

Note: In bold type: significant differences between the OSAS-types.
Abbreviations: A, Adenoids; E, Epiglottis; L, Larynx; N, Nose; O, Oropharynx; T, Tongue; V, Velum.

The nose, adenoids, velum, oropharynx, tongue, epiglottis, larynx (NAVOTEL) score was created following these findings by adding three frequent obstructive sites in children: N for the nose, A for adenoids, and L for the larynx. The scoring system is identical to VOTE: 0 in the case of normal patency, 1 for partial obstruction, and 2 for complete obstruction. The VOTE (out of 8) and NAVOTEL (out of 14) scores were calculated for each patient; their scores were harmonized for comparison.

3.3 | Correlation between the scoring systems and OAHI

The correlation between VOTE scores and the OAHI was insignificant: $\rho = 0.19$ confidence interval (CI) 95[−0.0037; 0.3768] $p = 0.054$. There was a correlation between NAVOTEL scores and OAHI: $\rho = 0.20$ CI 95 [0.005; 0.385]; $p = 0.044$. The correlation between the two scoring systems was significant: $\rho = 0.76$ CI 95 [0.66; 0.83] and $p < 0.001$. No significant differences were found according to the OSAS type. The relationship between OAHI and NAVOTEL scores remained significant in a multivariate analysis adjusting for age ($p = 0.04$). When NAVOTEL increased by 1 unit, the OAHI increased on average by 0.99; $p = 0.04$.

3.4 | Therapeutic consequences: Clinical examination, VOTE, and NAVOTEL

A total of 123 surgical procedures were performed to treat OSAS in the series (Figure 1). Seventy-three surgical actions were indicated by clinical examination: 51 tonsillectomies on endobuccal examination for hypertrophy, 14 adenoidectomies on fiberoptic laryngoscopy, 7 turbinoplasties, and 1 septoplasty. DISE with the VOTE scoring system indicated eight surgical actions: four tonsillectomies, three epiglottoplasties, and one lingual tonsil resection. DISE with

TABLE 3 Distribution of characteristics, obstruction sites, and VOTE and NAVOTEL scores by OSAS type.

		Type I (*n* = 65)	Type II (*n* = 14)	Type III (*n* = 20)	*n*	*p*
Age, mean (SD)		7.21 (3.71)	9.65 (3.61)	5.59 (3.05)	99	**<0.001**
Gender	M	40 (62%)	3 (21%)	9 (45%)	52	**0.018**
	F	25 (38%)	11 (79%)	11 (55%)	47	–
OAHI, mean (SD)		6.10 (7.47)	6.00 (4.61)	8.37 (10.3)	99	0.38
Patients with obstruction (%)	N	45%	50%	40%	99	0.36
	A	57%	79%	70%	99	**0.003**
	V	9%	29%	20%	99	**0.002**
	O	49%	57%	80%	99	**<0.001**
	T	32%	57%	60%	99	**<0.001**
	E	22%	29%	40%	99	**0.02**
	L	5%	0%	10%	99	0.18
No. of sites, mean (SD)		3.89 (0.954)	4.71 (1.14)	4.70 (1.08)	99	**<0.001**
VOTE score, mean (SD)		1.66 (1.03)	2.43 (1.50)	2.90 (1.37)	99	**<0.001**
NAVOTEL score, mean (SD)		3.20 (1.43)	4.07 (1.82)	4.60 (1.85)	99	**<0.001**

Note: Type I OSAS—isolated lymphoid tissue hypertrophy; type II—obese patient (BMI > 95th percentile); type III—patient with a craniofacial malformation or a neuromuscular disorder. In bold type: significant differences between the OSAS-types.

Abbreviations: A, adenoids; E, epiglottis; L, larynx; N, nose; O, oropharynx; OAHI, Obstructive Apnea-Hypopnea Index; OSAS, obstructive sleep apnea-hypopnea syndrome; T, tongue; V, velum.

NAVOTEL scoring system indicated 42 more surgical actions to VOTE: 25 adenoidectomies, 15 turbinoplasties, and 2 aryepiglottic fold sections with corniculectomy (classified as L2). The NAVOTEL scoring system did not omit any of the surgical indications.

Nine children (9%) in the series had a VOTE score of 0. Of these, the NAVOTEL scoring system found an obstructive site in 6 (66%), with 4 (44%) complete obstruction scored 2/2, and 3 (33%) multisite obstruction. Six surgical acts were performed on these patients: two supraglottoplasties, three adenoidectomies, and one turbinoplasty.

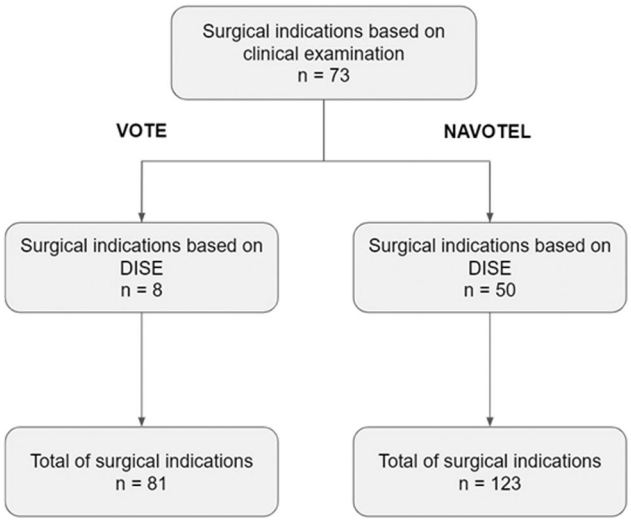

FIGURE 1 Comparison of surgical indications according to VOTE and NAVOTEL scoring systems. DISE, drug-induced sleep endoscopy.

4 | DISCUSSION

DISE in children highlighted that three frequent obstructive sites were not included in the VOTE scoring system. This led to the proposal of the NAVOTEL scoring system, which is easy to use, allows to establish all surgical indications, is significantly correlated with the severity of the OAHI ($p = 0.2$; $p = 0.04$) and identifies an obstructive site in most patients with a VOTE score of 0.

To our knowledge, the number of 99 patients is one of the largest in the literature. Patients without preoperative sleep recordings were nonincluded, primarily infants and patients with complex neurological pathologies, for whom getting a reliable recording is difficult. The 57 patients who underwent DISE without preoperative sleep recording are the object of another study. Their noninclusion was necessary for the homogeneity of this study conducted on patients with a documented OSAS.

Most sleep recordings were performed by respiratory polygraphy, not polysomnography, which remains the reference examination. This is explained by the long access time to polysomnography and the need to treat these children quickly. When the respiratory polygraphy is pathological, the OAHI is underestimated compared with that calculated by polysomnography.[16] The lack of standardization of preoperative registration is a weakness of this study, but it

would not have been possible to justify an extension of the time to management to obtain systematic polysomnography. In our practice, polysomnography is requested when respiratory polygraphy is normal, and the clinical suspicion of OSAS is high or when a failure of respiratory polygraphy is anticipated. This limitation is also a possible explanation for the low, albeit significant, correlation between the OAHI and the NAVOTEL score. Even if the OAHI does not reflect the full complexity of sleep pathology, it remains the main parameter used in the literature and clinical practice.

Awake fiberoptic laryngoscopy was not always performed despite the recommendations[20] for two reasons. First, this study was conducted during the Covid period, with a restriction on using fiberoptic laryngoscopy. Second, for some patients, when tonsillectomy was proposed on the oral examination, the fiberoptic laryngoscopy was performed in the operative room without modification of the indication for general anesthesia.

For three patients, no obstructive sites were identified by DISE. Two hypotheses can explain this: either the anesthetic protocol did not recreate the sleep conditions during which the apneas occur, or the sleep recording was performed during an episode increasing the obstruction and the OAHI, for example, during a nasopharyngeal infectious episode.

The distribution of obstructive sites differs according to the patient's medical history. Patients with type II and III OSAS had significantly more obstructive sites than type I: 4.7 versus 3.9 ($p < 0.001$). Type III OSAS patients presented oropharyngeal obstruction in 80% of the cases and a tendency to laryngeal obstruction (10% vs. 5% and 0%). A more frequent adenoid and tongue base obstruction was noted in type II and III OSAS, possibly due to an enlargement of the tonsil lymphoid tissue. There are little data in the literature regarding the distribution of obstructive sites according to the patient's medical history.[21–24] Hyzer et al. found that obese children with Down's syndrome had more oropharyngeal obstruction due to enlarged tonsils.[21] Maris et al. reported an infrequent involvement of the tongue base (24%).[23] The literature also highlights a higher prevalence of sleep laryngomalacia in children with Down syndrome.[25] In children with type II OSAS, Hyzer et al.[21] found more frequent oropharyngeal obstruction but less due to the tongue base compared to children with type I OSAS, which is in line with our observations for the oropharyngeal involvement but not for the tongue base, which was more frequent in type II OSAS (57% vs. 32%).

Regarding multisite involvement, our series reveals frequent multisite obstruction in children with type II or III OSAS compared to type I and a significantly higher NAVOTEL score. This may be explained by hypertrophy of lymphoid tissue aggravating OSAS in patients who already have reduced airways due to craniofacial malformation, hypotonia, or fatty infiltration. Blanc et al.,[26] in a study including 31 patients, did not find more obstructive sites in children with type III OSAS compared to type I (2.0 vs. 1.8; ns) or a statistical correlation between the preoperative OAHI and the number of obstructive sites. Hyzer et al.[21] found a trend toward more frequent multisite involvement for children with Down syndrome, compared to children with type I OSAS. The limitation of these findings was the consideration of only total obstructions in their definition of multisite involvement.

Nasal obstruction contributes to upper airway obstruction and OSAS in children.[27,28] In our series, 80% of children presented nasal or nasopharyngeal obstruction. Fitzpatrick et al.[29] found higher airway resistance during sleep with strictly oral breathing and suggested that the impact may be more significant in children due to smaller absolute dimensions.

Five children had sleep laryngomalacia, which manifests mainly after the age of 2 years by snoring and signs suggestive of sleep apnea. In contrast to awake laryngomalacia, it does not present with stridor, feeding difficulties, breathing difficulties, or weight loss. The prevalence in children with OSAS is approximately 3.9%, consistent with our data.[30] Sleep laryngomalacia is often associated with other obstructive sites.[31] DISE plays a fundamental role in the diagnosis: the collapse occurs during sleep when muscle tone is relaxed. Supraglottoplasty is the leading surgical management. In a series of 22 patients aged 2–17, Chan et al.[32] observed OAHI improvement following epiglottoplasty in 91% of patients.

Wilcox et al.[14] describe the existence of six scoring systems: VOTE, SERS, Chan, Bachar, Fishman, and Boudewyns. The VOTE[13] was developed based on the experience of over 8000 adult DISEs and designed as a surgical planning tool for adults with OSAS by locating and characterizing the most found obstructive sites. It was not intended to be used as a disease assessment or prognostic tool and has not been validated for this purpose. Nevertheless, it is widely used in adults and children.[14] It ignores common obstructive sites in children, such as the nasal cavity and adenoids, and specific laryngeal pathologies, such as sleep laryngomalacia. In our series, VOTE incorrectly classified six children as unaffected and considered only 8 of the 50 procedures added by DISEs. Chan DK et al.[33] developed in 2014 a scoring system that consists of five sites with the addition of the adenoids and the replacement of the E of VOTE by the supraglottic larynx. It does not consider nasal obstruction and groups epiglottic and laryngeal obstructions. It is also more complex, with four levels of obstruction. The authors did not find a statistically significant with OAHI. In 2016, Lam DJ et al.[34] created the Sleep Endoscopy Rating Scale (SERS), which uses three levels of obstruction and adds the nasal cavity to Chan's score. The epiglottis and the tongue base are grouped into “hypopharynx,” and the larynx appears in “arytenoids.” The authors found a statistically significant correlation with the OAHI. Merging two sites of obstruction into the “hypopharynx” does not seem relevant: the surgical treatments are different, and these two sites are not necessarily associated. Boudewyns et al.[35] scoring system, created in 2017, is adapted to a population under 2 years of age, includes four degrees of obstruction, with six possible sites of obstruction (adenoids, tonsils, tongue base, palate, epiglottis, laryngomalacia) characterized as fixed or dynamic, in addition to a general assessment of hypotonia. This score omits nasal obstruction, which is essential in infant ventilation. Fishman et al.[36] assessed the upper airways in five subsites (nasal cavity, nasopharynx, lateral walls of the oropharynx, tongue base, and supraglottis). They considered the primary site of obstruction

identified and its severity in four grades. The epiglottis and the soft palate, frequently responsible for obstruction in type II and III OSAS, are missing. Bachar et al.[37] classification, created for adults, scores with the letters NPTLH for "nose, palate, tongue, larynx, hypopharynx." This score does not differentiate the nasal cavity from the nasopharynx, nor the soft palate from the tonsils. This makes it unsuitable for pediatric practice because it does not correspond to the obstacles encountered or the possible surgeries. A seventh score, not cited by Wilcox et al., has been developed by Williamson et al.[38] It is adapted to pediatric practice and is exhaustive in terms of analysis of obstructive sites, with 10 different levels. However, this completeness makes it more challenging to implement and risks overrepresent some obstructive sites. For example, the vallecula and the lingual tonsils are two different sites, as well as the aryepiglottic folds and the arytenoids.

5 | CONCLUSION

This study allowed a better understanding of the distribution of obstructive sites responsible for OSAS in children. It allowed the creation of the NAVOTEL scoring system, which is simple to use while allowing the exhaustiveness necessary for pediatric DISE. It is statistically correlated with the OAHI severity. We use it in daily clinical practice to optimize the management of our patients. NAVOTEL scoring system may replace VOTE in children to record the results, improve communication between practitioners, optimize therapeutic management, and homogenize publications on pediatric OSAS.

AUTHOR CONTRIBUTIONS

Qarbal Jawad: Data collection; statistical analysis; data presentation; writing of the original draft. **Claire Le Treut-Gay**: Conceptualization; data collection; draft revision. **Allali Laure**: Conceptualization; data collection; draft revision. **Rossi Marie-Eva**: Conceptualization; data collection; draft revision. **Nicollas Richard**: Conceptualization; resources; validation. **Moreddu Eric**: Conceptualization; methodology; project administration; resources; validation; draft revision and writing of the final version of the manuscript.

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Not Applicable.

CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request. Data are available, accessible, discoverable, and usable.

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