Development and validation of the international pediatric sleep endoscopy scale (IPSES)

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Abstract

Objectives: To develop and validate a consensus international pediatric sleep endoscopy scale (IPSES) for pediatric drug-induced sleep endoscopy (DISE).

Methods: Existing published DISE ratings scales were reviewed in order to develop a consensus rating scale synthesizing the most common features and adding new elements to address areas of controversy. Samples of 30 de-identified DISE video recordings were reviewed to develop and refine the scale. After the consensus scale was defined, a separate sample of 25 de-identified DISE videos were scored with the new consensus scale by the development group and a panel of independent raters. A weighted kappa statistic was used to quantify the inter-rater and intra-rater reliability of the consensus scale at each anatomic level.

Results: Among all raters, intra-rater reliability was most variable for the nasal airway (kappa range 0.33-0.94) and best for the lateral oropharynx (kappa range 0.68-0.95). Inter-rater reliability ranged from 0.43 for the nasal airway to 0.57 at the soft palate.

Conclusion: The IPSES is a reliable consensus scale that reflects the most common features of existing scales and can be adopted as a universal scoring scale for pediatric DISE.

Keywords: DISE, sleep endoscopy, consensus, pediatric, sleep apnea, validation, reliability

1.1 Introduction

Drug-induced sleep endoscopy (DISE) has gained increasing popularity in the evaluation of obstructive sleep apnea (OSA) in children in recent years.[1-3] Though it has proved to be a useful tool in helping to identify sources of obstruction and formulate treatment plans, DISE has suffered from a lack of standardization in scoring or documentation of findings.

Many different scoring systems have been proposed in recent years, and though these systems share many similarities,[4,5] there is no scoring system that has been universally accepted or adopted. This has prompted recent efforts by the International Pediatric Otolaryngology Group (IPOG) to bring together a working group comprised of experts in pediatric DISE from around the world to generate a consensus statement regarding the elements and scoring rubric that should form the basis of a consensus rating scale. A parallel effort, led by a subset of this working group had already been working on just such a consensus rating scale at the time that the IPOG group was assembled, and the two efforts were dovetailed to promote universal agreement and acceptance.

Thus, the aim of this project was twofold: to create and validate a straightforward DISE grading scale for children by adopting the common elements from the existing published pediatric DISE rating scales.

2.1 Methods

This was a cross-sectional validation study using existing DISE recordings. Recordings were drawn from children ages 2-18 years who underwent DISE at any time from 2015-2022 for the treatment of OSA at one of four academic centers (Oregon Health and Science University, Portland OR, Children's Hospital Colorado, Aurora CO, the Chinese University of Hong Kong, Hong Kong SAR, China, and the Antwerp University Hospital, Antwerp, Belgium). To improve generalizability, the videos reflected a variety of comorbidities and past surgical history. Some patients were surgically naïve, while others were post-tonsillectomy. No restriction was made based on comorbidity. Recordings were selected by the development group (DL, NF, KC, AB) based on video quality and completeness of the examination (i.e. showing all portions of the upper airway from the nasal cavity to the supraglottis) with sufficient clarity to allow a meaningful rating throughout (e.g. not obscured by excessive secretions). No other specific selection criteria were defined, but an effort was made to collect videos representing a diverse group of patients and patterns of obstruction. All videos were edited to remove lengthy or redundant segments showing the same anatomic level with the same pattern of obstruction down to <2 minutes in length to facilitate efficiency in review and to remove identifying features.

2.2 International Pediatric Sleep Endoscopy Scale (IPSES) Development

The development group met virtually every 6 to 8 weeks to discuss existing rating scales and areas of commonality and controversy. Attention was paid to the nuances of scoring at each anatomic level and challenging patterns to describe or score. Based on these discussions, a proposed consensus rating scale was formulated, with sample de-identified still pictures taken from previously recorded DISE videos to illustrate and define the scoring rubric. In addition, a scoring form (Fig 1) was developed without detailed descriptions or pictures, as a template for scoring to be used either in paper or electronic format in a clinical setting. A development set of videos were collected. Each member evaluated the DISE video and completed the scoring form. Several rounds of independent scoring by each author using a sample of 30 de-identified videos were done, with comparison and extensive discussion of areas of scoring disagreement in order to further refine and standardize the rating scale (Fig 2).

The final scoring rubric is identical to the one adopted by the International Pediatric Otolaryngology Group (IPOG) consensus on scoring of pediatric DISE.[6] In brief, there are 3 categories for severity of obstruction: 0: non obstructive- <50% obstruction, 1: partial – 50-90% obstruction, 2: complete- > 90% obstruction. The severity of collapse at each anatomic level is determined by the maximum degree of obstruction observed. These ratings were applied at 7 anatomic levels in the upper airway: the nasal airway (from nostrils to choana, including assessment of inferior turbinates and septum), nasopharynx (assessment of adenoid obstruction), soft palate, lateral oropharynx (including palatine tonsils if present), tongue base (including lingual tonsils if present), epiglottis (distinct from glossoptosis or tongue base obstruction), and arytenoids (degree of anterior prolapse with glottic obstruction).

Beyond the IPOG scoring rubric, some novel features were developed that are unique to the newly developed rating scale. These features are intended to either help clarify challenging areas to assess (e.g. nasal airway and epiglottic obstruction) or facilitate systematic documentation of qualitative description (i.e. the pattern of obstruction and fixed vs dynamic collapse).

2.2.1 Nasal airway assessment –Each side of the nasal airway is graded independently. These ratings can be reported separately or combined in a single rating of nasal obstruction for ease of reporting. A combined nasal obstruction score can be defined as: non-obstructive = both sides scored 0, partial = at least one side scored ≥ 1), complete = both sides scored 2.

2.2.2 Clear definition of epiglottic collapse – In order to clearly differentiate epiglottic retroflexion that is secondary to glossoptosis vs retroflexion and obstruction that is independent of the tongue base, we chose to define the former as non-obstructive with respect to the epiglottis, and only assessed epiglottic collapse based on the degree to which the epiglottis collapses and obstructs *independent* of glossoptosis or tongue base obstruction.

2.2.3 Description of obstructive pattern at each anatomic level –To systematically capture a description of the pattern of collapse at the anatomic levels where dynamic movement is common (velum, lateral pharyngeal walls, tongue base, and epiglottis), different patterns were broadly grouped into two or three categories indicating the direction of movement from maximum opening to maximum closure (e.g. anterior-posterior, lateral, circumferential).

2.2.4 Assessment of fixed vs dynamic collapse –At sites where dynamic movement is common, a designation of fixed obstruction (no movement observed) or dynamic (at least some movement present) was made, with the assessment of severity of obstruction based on the maximal degree of obstruction observed.

2.3 IPSES Validation

An independent sample of 25 DISE videos was collected, distinct from the original videos used in scale development. This validation set of videos was again compiled from previously recorded DISE videos with contributions from all members of the development group, edited for length and anonymity as needed.

All videos were scored by all reviewers on two different occasions separated by at least one week to allow calculation of both inter-rater and intra-rater reliability. A linear-weighted Cohen's kappa statistic was calculated to assess the reliability of the ratings of severity of obstruction (non-obstructive, partial, severe) at each of the 7 anatomic levels. Of note, because other features of the rating scale were considered descriptive in nature (e.g. pattern of obstruction, fixed vs dynamic, etc) and not intended to be quantified, reliability testing was not calculated for these more granular features. To assess for external validity, a second group of independent raters was recruited (EK, DS, PB) and asked to review all videos using only the IPSES scoring guide and accompanying scoring form (Figs 1 and 2). This was intended to reflect the same information that a reader of the current manuscript would have in scoring an unfamiliar DISE video. To summarize the obstructive patterns represented in the validation set of videos, the initial scores (0, 1, 2) from all reviewers were used to calculate mean obstructive scores at each anatomic site for each sample video.

3.1 Results

DISE videos from 25 patients were included in the final validation. Patient characteristic data were only available for 17 of these 25 due to IRB restrictions at a single institution (Table 1). Data from the patients who were not included are similar to those who were reported. Mean and median age were 8.6 and 7.2 years respectively, with a range of 2.5 to 17.9 years. A slight majority were male (53%), and approximately half were Caucasian. The majority of included patients were overweight (18%) or obese (35%), and the most common comorbidity was Down syndrome (23%). Mean IPSES scores for each video of the validation set are summarized in Figure 3 and demonstrate broad variability of obstructive pattern.

3.2 Intra-rater reliability

Intra-rater reliability varied considerably across raters and anatomic sites (Table 2). The site with the most variable intra-rater reliability was the nasal airway, ranging from fair (kappa 0.33) to almost perfect (kappa 0.94) while the lateral oropharynx was most reliable, ranging from substantial (kappa 0.68) to almost perfect (kappa 0.95).

3.3 Inter-rater reliability

Inter-rater reliability was less variable than intra-rater reliability, but also less reliable across all anatomic sites, as expected. When considering reliability across all raters, inter-rater reliability was moderate at all sites, with kappa ranging from 0.43 for the nasal airway to 0.57 at the soft palate (Table 3). The soft palate, oropharynx, tongue base, and arytenoids were all similar in reliability (kappa 0.55-0.57). There was a substantial difference in inter-rater reliability when considering ratings of the development group vs the external raters. The development group demonstrated substantial agreement at the soft palate, tongue base, epiglottis, and arytenoids (kappa 0.62-0.73) and no worse than moderate agreement at other levels (kappa 0.48-0.52). However, the independent raters showed only fair reliability for ratings of the nasal airway (0.30-0.38) and epiglottis (0.23) and moderate to substantial reliability for all other levels (0.49-0.62).

4.1 Discussion

With the growing popularity of DISE in the evaluation and treatment of pediatric OSA, it has become increasingly important to standardize the assessment and documentation of DISE findings. In this study, we developed and validated a semi-novel rating scale for pediatric DISE that includes a scoring rubric for severity of obstruction which is identical to the one agreed upon by the IPOG DISE working group. The purpose of this effort was to synthesize and optimize the most commonly reported elements and scoring methods in studies using pediatric DISE so as to propose and validate a consensus rating scale that can be universally adopted moving forward. Our data demonstrate that the IPSES has moderate-excellent intrarater reliability and moderate interrater reliability which is comparable to previously published DISE scales.[7-11] The interrater reliability was slightly worse in the nasal airway (kappa 0.43-0.46) and slightly better for the soft palate, oropharynx, and tongue base (kappa 0.56-0.57).

Previously published rating scales include the Fishman scale[7], Chan-Parikh scale[8], the SERS[11], the Williamson scale[10], the VOTE,[12] and others[9,13-16]. All have shown similar degrees of reliability and, where tested, correlation with OSA severity. It is striking how similar these scoring systems are in terms of the scoring rubrics, structures assessed, and overall scheme. All are based on categorical ratings of three or four categories of obstructive severity at multiple locations in the upper airway. Unlike the VOTE, commonly used in adults, pediatric DISE requires consideration of additional anatomic levels that can contribute to obstruction in children (i.e. nasal airways, adenoids, and arytenoids). These sites are specifically recommended to be included in any pediatric DISE assessment in the consensus statements from the AAO-HNSF[1] and IPOG.[6] Studies describing pediatric DISE findings often supplement or adapt published scoring systems, and the adaptations tend to be similar. For example, some studies have modified the CP scale to include ratings of the nasal airway,[17,18] while others that use VOTE scoring also include ratings of the adenoids and supraglottis.[2] Finally, some authors have reported on DISE-directed interventions without using a specifically defined scoring system, but typically with elements very similar to what we have described.[14,19,20] Taken together, these reports reflect a de facto consensus in the anatomic areas and features that are clinically relevant and important to score. We believe that the IPSES scale described in this study combines the best features of these existing scales in a form that reflects this de facto consensus and strikes the best balance of granularity and clinical usability. It can be used in both the surgically naïve and in those who have had previous surgery for sleep apnea like adenotonsillectomy. It also closely reflects the IPOG consensus statement describing the elements that should be included in a consensus pediatric DISE scale.[6]

It is important to note that the IPSES includes more granularity in the grading of DISE videos than the IPOG consensus statement describes. This may facilitate identification of specific anatomic endotypes and ultimately enable a more personalized approach to treatment.

The IPSES expands beyond the fundamental scoring rubric in four ways: nasal airway assessment, description of epiglottic collapse, pattern of obstruction, and whether the obstruction is dynamic or fixed. Since nasal obstruction can be either unilateral or bilateral, the decision was made to assess both sides of the nasal airway independently. The nasal airway has often been overlooked in past assessments of the upper airway during sleep, but it is clear that nasal breathing is important in maintaining a patent upper airway during sleep and that nasal obstruction can contribute to sleep apnea in children.[1,21,22] With respect to the epiglottis, obstruction that is independent of the tongue base is a distinct endotype that could be addressed separately from the tongue base such as with an epiglottopexy.[23,24] Though not reflected in the numerical categorical ratings at each level, capturing some description of the pattern of obstruction at each level in a systematic format (rather than simply by a narrative qualitative description) was felt to be important in helping to guide clinical decision-making and may prove useful in determining predictors of outcome. This concept is best illustrated by the consideration of concentric collapse at the soft palate as an important predictor of hypoglossal nerve stimulator candidacy in adults.[25] It was noted that some anatomic levels (soft palate, lateral oropharynx, tongue base, epiglottis, and arytenoids) can have obstruction that is either fixed (no observed movement), or dynamic, with clear obvious movement resulting in intermittent obstruction. This may reflect variation in plane of anesthesia as much as patient anatomy and may be an indirect measure of study quality (i.e. lack of dynamic movement might suggest an inadequate study, though this has not been clearly demonstrated).

Our study has important limitations. First, the videos were collected retrospectively from the course of routine clinical practice and selected primarily for video quality and completeness of the assessment, without specific inclusion and exclusion criteria in mind. As such they included patients with variable surgical histories and comorbidities, and it is possible that there was selection bias present where specific patterns of obstruction may be over-represented relative to an unselected population. However, Figure 3 demonstrates broad variability with respect to the patterns of obstruction among the sample videos. One of the priorities of the development group was to include videos demonstrating patterns of obstruction that might be challenging to rate with existing systems, for example, lateral epiglottic collapse or lateral tongue base collapse. This may have negatively impacted both inter and intra-rater reliability. However, we believe this also increases generalizability and better reflects real-world practice conditions. Second, all raters included in this study were experienced DISE practitioners, and it is possible that the reliability among novice or occasional DISE users could be worse than what we found. However, this could be true of any DISE rating scale. Familiarity with the rating scale may also account for the slightly worse interrater reliability among the external raters relative to the development group. If this scale can be universally adopted and regularly used, greater familiarity should lead to greater reliability. Strengths include the inclusion of DISE practitioners from different countries and institutions, the use of de-identified videos, and the blinded review by an independent group of raters, separate from the development group. An additional strength is the deliberate attempt to clarify areas of scoring controversy that have not previously been addressed.

5.1 Conclusion

The IPSES consensus rating scale for pediatric DISE utilizes the IPOG endorsed scoring rubric as the foundation for a more granular scale. With similar reliability to existing rating scales, we would advocate for the IPSES to be adopted as a universal pediatric DISE rating scale in order to facilitate outcomes research for complex pediatric OSA and multi-institutional collaboration.

Tables

Table 1. Patient Characteristics*

	N (%)
Age at DISE (years)	
Mean	8.6 ± 4.0
Median [IQR]	7.2 [6.0, 10.8]
Female	8 (47)
Race	
Caucasian	9 (53)
Asian	6 (35)
Black	1 (6)
Hispanic	1 (6)
Weight Category	
Normal Weight	8 (47)
Overweight	3 (18)
Obese	6 (35)
Comorbidities	
None	8 (47)
Down syndrome	4 (24)
Neuromuscular disorder	2 (12)
Craniofacial anomaly	1 (6)
Other	2 (12)
Past Surgical History	
None	11 (65)
Adenotonsillectomy	5 (29)
Palatoplasty, mandibular distraction	1 (6)

*Patient characteristic data only available for 17 of 25 patients due to IRB restrictions from one institution

Table 2. Intra-rater Reliability

	Rater 1	Rater 2	Rater 3	Rater 4	Rater 5	Rater 6	Rater 7
Anatomic Level	Cohen's Kappa	Cohen's Kappa	Cohen's Kappa	Cohen's Kappa	Cohen's Kappa	Cohen's Kappa	Cohen's Kappa
NA – R	0.46 [0.17, 0.75]	0.75 [0.53, 0.97]	0.80 [0.60, 0.99]	0.53 [0.25, 0.82]	0.33 [0.04, 0.62]	0.90 [0.76, 1.00]	0.94 [0.81, 1.00]
NA – L	0.56 [0.22, 0.89]	0.81 [0.63, 0.99]	0.85 [0.68, 1.00]	0.36 [-0.07, 0.79]	0.49 [0.19, 0.79]	0.79 [0.59, 0.98]	0.85 [0.68, 1.00]
NP	0.60 [0.30, 0.90]	0.75 [0.47, 1.00]	0.80 [0.60, 1.00]	0.78 [0.54, 1.00]	0.44 [0.21, 0.66]	0.94 [0.80, 1.00]	0.69 [0.41, 0.93]
V	0.71 [0.48, 0.95]	0.95 [0.86, 1.00]	1.00 [1.00, 1.00]	0.51 [0.24, 0.78]	0.62 [0.37, 0.87]	0.95 [0.82, 1.00]	0.80 [0.59, 1.00]
LW	0.87 [0.68, 1.00]	0.68 [0.44, 0.91]	0.77 [0.53, 1.00]	0.74 [0.54, 0.94]	0.91 [0.75, 1.00]	0.91 [0.74, 1.00]	0.95 [0.86, 1.00]
TB	0.74 [0.53, 0.95]	0.66 [0.46, 0.86]	0.91 [0.78, 1.00]	0.85 [0.69, 1.00]	0.38 [0.09, 0.66]	1.00 [1.00, 1.00]	0.95 [0.86, 1.00]
EP	0.79 [0.59, 0.98]	0.70 [0.41, 1.00]	0.96 [0.87, 1.00]	0.60 [0.28, 0.92]	0.38 [0.09, 0.66]	1.00 [1.00, 1.00]	0.95 [0.84, 1.00]
AR	0.86 [0.57, 1.00]	0.81 [0.63, 1.00]	0.87 [0.72, 1.00]	0.72 [0.35, 1.00]	0.78 [0.62, 0.94]	1.00 [1.00, 1.00]	0.91 [0.78, 1.00]

Kappa Interpretation	0.0-0.2: Slight	0.2-0.4: Fair	0.4-0.6: Moderate	0.6-0.8: Substantial	0.8-1.0: Almost perfect

Table 3. Inter-rater reliability

	Development Group	Independent Raters	All raters (7)
Anatomic Level	Cohen's Kappa	Cohen's Kappa	Cohen's Kappa
NA – R	0.56 [0.36, 0.77]	0.30 [0.13, 0.47]	0.43 [0.28, 0.59]
NA – L	0.48 [0.28, 0.68]	0.38 [0.18, 0.59]	0.46 [0.29, 0.63]
NP	0.52 [0.32, 0.72]	0.49 [0.24, 0.74]	0.50 [0.33, 0.67]
V	0.65 [0.47, 0.83]	0.49 [0.25, 0.72]	0.57 [0.39, 0.74]
LW	0.52 [0.34, 0.71]	0.62 [0.39, 0.86]	0.56 [0.40, 0.71]
TB	0.68 [0.52, 0.84]	0.49 [0.26, 0.71]	0.56 [0.40, 0.72]
EP	0.73 [0.56, 0.91]	0.23 [-0.03, 0.49]	0.50 [0.34, 0.65]
AR	0.62 [0.44, 0.81]	0.49 [0.25, 0.74]	0.55 [0.38, 0.72]

Kappa Interpretation	0.0-0.2: Slight	0.2-0.4: Fair	0.4-0.6: Moderate	0.6-0.8: Substantial	0.8-1.0: Almost perfect

Figure Legend

Figure 1. IPSES Scoring Template

Figure 2. IPSES Scoring Guide

Figure 3. Mean IPSES Scores For Each Sample DISE Video
NAR = Nasal airway right, NAL = Nasal airway left, NP = Nasopharynx, V = Velum, LW = Lateral Wall/Tonsils, TB = Tongue Base, EP = Epiglottis, AR = Arytenoids

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