Pediatric Drug-Induced Sleep Endoscopy: Technique and Scoring System

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Abstract

Drug-induced sleep endoscopy (DISE) is an invaluable tool for identifying sites of obstruction for patients with obstructive sleep apnea (OSA). During DISE, the patient is in a state of drug-induced sleep, and a flexible laryngoscope is passed through the nose into the upper airway. Sites of obstruction are visualized and scored to guide surgical management. Currently, there is no universally accepted method of DISE analysis and scoring. This limitation in comparability impedes large-scale analysis between clinicians, institutions, and studies. In this report, we propose a standardized method of scoring and performing DISE in children with OSA. Our DISE scoring system is internally developed, consistent through the study, and addresses all levels of potential upper airway obstruction.

Introduction

Obstructive sleep apnea (OSA) is a common childhood syndrome that can be associated with significant morbidity and is known to be detrimental to neurobehavioral, cardiovascular, endocrine, and metabolic health and development in children [1]. The prevalence of OSA is between 1% and 4%, however, it is likely under-diagnosed and under-treated [2,3]. The diagnostic gold standard for OSA is overnight polysomnography (PSG); however, this does not provide anatomic information related to the cause of airway obstruction.

Drug-induced sleep endoscopy (DISE) in children is a useful tool for identifying sources of obstruction and planning future surgical intervention. During this procedure, the patient is in a state of drug-induced sleep, and a flexible laryngoscope is passed through the nose into the upper airway. Sites of obstruction are visualized and documented, and this information is used to guide surgical management. In general, DISE demonstrates statistical promise in terms of safety, validity, test-retest reliability, and inter/intra-rater reliability [4-7]. The purported indications for DISE in children include documented persistent OSA after tonsillectomy and adenoidectomy, high risk for persistent OSA after tonsillectomy and adenoidectomy, significant sleep-disordered breathing (SDB) or OSA in patients with small tonsils and adenoids, concern for occult or sleep-state dependent laryngomalacia, and to confirm the level of obstruction prior to placement of hypoglossal nerve stimulator [5].

There are several published scoring systems, however, none are used universally. The lack of consensus prevents objective outcome analysis between clinicians, institutions, and studies [8]. This limitation in comparability is important to consider as it may impede large-scale analysis such as systematic reviews and meta-analyses of refractory OSA patients. Commonly used DISE scoring systems differ in how many sites of obstruction are evaluated, description of airway narrowing, and obstruction configurations [5,9]. In this report, we propose a standardized method of scoring and performing DISE in children with refractory OSA. This comprehensive and easy to follow method takes into account all potential sites of obstruction, degree of narrowing, and obstruction configuration.

Technical Report

DISE is performed in the operating room with a pediatric anesthesia team that provides the necessary sedation. Following standard protocol for pediatric patients, anesthesia is first induced using an inhaled anesthetic agent allowing for intravenous access to be obtained. The inhalation agent is then discontinued and a propofol infusion dosed appropriately for the child’s age and weight is used for the remainder of the procedure. The patients are monitored closely while placed in supine position with the chin in neutral position.

DISE is initiated when audible or palpable snoring is noted. The flexible fiberoptic laryngoscope is passed...
into the nose bilaterally. The flexible laryngoscope is advanced into the nasopharynx, followed by the oropharynx, and ends at the level of the supraglottic larynx. The airway patency is evaluated throughout the exam. The DISE procedures are recorded in video form and still images are taken during the procedure for later review and documentation (Figure 1). The DISE procedure is documented using a uniform template and scoring system (Figure 1). The scoring system records the operator’s evaluation of the bilateral nasal airway, adenoid, retropalatal airway, oropharyngeal airway, retrolingual airway, and laryngeal airway separately. Obstruction at each site or by the specified structure is broadly categorized as non-obstructive, partially obstructive, or significantly obstructive (Figure 1). The completed template results in a narrative record of the DISE findings for that case.

| Site                          | No Obstruction                                                                 | Partial Obstruction                                         | Significant Obstruction                                    |
|-------------------------------|--------------------------------------------------------------------------------|-------------------------------------------------------------|-----------------------------------------------------------|
| Nasal Airway†                 | Airway is patent at anterior turbinates.                                       | Anterior turbinate contacts suprarnasal mucosa.             | Anterior turbinate contacts suprarnasal mucosa.           |
| Adenoid                       | Less than 25% of oropharynx blocked or unobstructed.                           | 25–75% of oropharynx blocked                                | More than 75% of oropharynx blocked                       |
| Palate                        | No movement in respiratory cycle, no obstruction.                              | Airway movement causing partial obstruction.                | Completely closed oropharynx                              |
| Tonsils and Lateral Pharyngeal Walls | Long less than 25% airway obstruction.                                         | 25–75% obstruction of airway.                              | More than 75% obstruction of airway.                      |
| Tongue Base                   | White points retrognathic area, no movement during separation.                | Pale, white points partially or completely but origin is well posterior. | Pale points anteriorly with flap epiglottis reaches posterior pharyngeal wall. |
| Lingual Tonsil                | Not muffled or absent.                                                         | Pale, white points partially or completely but origin is well posterior. | Pale points anteriorly with flap epiglottis reaches posterior pharyngeal wall. |
| Vallecula†                    | Pale, white points partially filled by lingual tonsil.                         | Vallecula partially filled by lingual tonsil at tongue base. | Vallecula completely filled by lingual tonsil at tongue base |
| Epiglottis                    | Normal, evenly white points.                                                   | Two-point contact with posterior pharyngeal wall and airway patent. | Two-point contact with posterior pharyngeal wall and airway patent. |
| Aryepiglottic Folds§          | True and false vocal cord visualized easily.                                   | Mild shortening, partially contacting epiglottis, vocal cord narrow. | Partial to no cords not seen as epiglottis pulled forward by space OC light. |
| Arytenoids                    | Vertical, no collapse.                                                        | Movement symmetric and mediolateral during separation.      | Complete collapse into glottis.                           |

**FIGURE 1: Drug-induced sleep endoscopy (DISE) scoring system**

This figure depicts the scoring system used with overlying picture examples. †Only left turbinate example pictures are provided, however, in practice the right and left nasal airway documented separately; ‡For vallecula example pictures, see tongue base and lingual tonsil rows. §Aryepiglottic folds were visualized with and without chin lift during DISE.
**Discussion**

The scope of DISE research is limited by lack of consistent quantitative data and range of methodology used among institutions and otolaryngologists. The adoption of a standard method and scoring system for pediatric DISE would help remedy this constraint on DISE research. The most obvious limitation is the lack of consistent scoring systems among studies and institutions. Six different scoring systems (Bachar, Boudewyns, Chan, Fishman, Sleep Endoscopy Rating Scale (SERS), Velum, Oropharynx and lateral pharyngeal walls, Tongue base and Epiglottis (VOTE)) have been used to report pediatric DISE findings (Table 1)[10-15]. The goal of all these scoring systems is to concisely and consistently communicate findings during DISE in a manner that can be quantified and studied. However, these systems are each unique with regard to the anatomic sites included in the system and in their manner of quantifying and characterizing airway.

| Scoring System | Sites Evaluated | Quantification of obstruction | Additional comments |
|----------------|-----------------|------------------------------|--------------------|
| Bachar et al. 2012 [10] | 1. Nasal airway and Nasopharynx | Score of 1 or 2 given for partial or complete obstruction respectively | This grading system converts to a NTPHL staging system. Sites without obstruction are not included in the staging for that patient. |
| | 2. Palate and tonsils | | |
| | 3. Tongue base | | |
| | 4. Hypopharynx | | |
| | 5. Larynx | | |
| Boudewyns et al. 2014 [11] | 1. Adenoid | Score of 0 for no obstruction, 1 for less than 50% obstruction, 2 for between 50-75% obstruction, and 3 for greater than 75% obstruction | This system includes a general impression of hypotonia as present or absent. |
| | 2. Palate | Score of 0 for no collapse, 1 for collapse present | |
| | 2. Tonsils and oropharynx | Score of 0 for no obstruction, 1 for less than 50% obstruction, 2 for between 50-90% obstruction, and 3 for tonsils that touch at midline. | |
| | 3. Tongue base | Score of 0 for no obstruction, 1 for partial obstruction, 2 for complete obstruction | |
| | 4. Epiglottis | Score of 0 for no collapse, 1 for collapse present | |
| | 5. Larynx | Laryngomalacia is noted to be absent (score of 0) or present (score of 1) | |
| Chan et al. 2014 [12] | 1. Adenoid | | |
| | 2. Velum | | |
| | 3. Lateral pharyngeal walls and oropharynx | Score of 0 for no obstruction, 1 for less than 50% obstruction, 2 for between 50-99% obstruction, and 3 for complete obstruction | |
| | 4. Tongue base | | |
| | 5. Epiglottis and | | |
| Supraglottis | 1. Nasal airway 2. Nasopharynx 3. Lateral pharyngeal walls and oropharynx 4. Tongue base 5. Epiglottis and supraglottis |
|-------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Score of 0 for no obstruction, 1 for mild obstruction, 2 for moderate obstruction and 3 for severe obstruction | This system includes severity of OSA (mild, moderate and severe), level of confidence in findings, and quality of examination (poor, fair and good). The interpreter is asked to determine the primary site of obstruction or select a combination of sites |

| SERS [14] | 1. Nasal airway 2. Nasopharynx 3. Velum 4. Lateral pharyngeal walls and oropharynx 5. Hypopharynx 6. Larynx |
|-----------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Score of 0 for no obstruction, 1 for partial obstruction, 2 for complete obstruction | |

| VOTE [15] | 1. Velum 2. Oropharynx and Lateral pharyngeal walls 3. Tongue base 4. Epiglottis |
|-----------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Score of 0 for no obstruction, 1 for partial obstruction, 2 for complete obstruction | This system characterizes the pattern of obstruction as anteroposterior, lateral or concentric |

| Proposed Scoring System | 1. Nasal airway 2. Adenoid 3. Palate 4. Tonsils and Lateral pharyngeal walls 5. Tongue base 6. Lingual tonsils 7. Vallecula 8. Epiglottis 9. Ayeroepiglottic folds 10. Arytenoids |
|------------------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Airway patency is generally categorized at each site to have no obstruction, partial obstruction, or significant obstruction | See Figure 1 for definitions of the obstructive categories at each site. A numeric scoring system could be applied to this system as needed for data collection with a score of 0 for no obstruction, 1 for partial obstruction, 2 for significant obstruction |
TABLE 1: Drug-induced sleep endoscopy (DISE) scoring systems used in children

| Scoring System | Description |
|----------------|-------------|
| VOTE           | Velum, Oropharynx and lateral pharyngeal walls, Tongue base and Epiglottis |
| SERS           | Sleep Endoscopy Rating Scale |
| OSA            | Obstructive sleep apnea |

The VOTE system is the most studied scoring system and has been used in both children and adults [15]. The system is concise and easy to use. However, a major limitation of the VOTE scoring system for use in children is the omission of the nasopharyngeal and supraglottic sites. The Chan scoring system is similar to the VOTE system but includes the supraglottis and lingual tonsils specifically, which are important sources of obstruction in children [12,15]. The Chan system uses a scoring system which correlates to the percentage of obstruction at all sites except the lingual tonsil which is described only as present or absent [12]. The Bachar system and SERS are unique in that they are an overall score of upper airway obstruction [10,14]. The Boudewyns system is unique in that it includes a designation of generalized hypotonia as present or absent and characterizes whether the obstruction is fixed or dynamic [11]. The Fishman system evaluates the degree of obstruction at several upper airway subsites but it is unique in that it also notes the quality of the exam, confidence in the findings, and OSA severity [13]. While the validity and reliability of DISE is promising, the clinical significance of these scoring systems is not well established [5-7]. Our DISE scoring system attempts to evaluate all levels of potential airway obstruction while maintaining a simple and easy to follow guide. The ideal scoring system should be simple and practical with proven reliability and should completely characterize the nature of obstruction in order to guide management.

Another concern with DISE research is how accurately intravenous anesthesia simulates normal sleep for the patient. It is important to note that all anesthetics used for DISE have some documented effect on sleep architecture [5,16]. The anesthetic-specific effect on the degree or level of obstruction has not been well studied. A combination of dexmedetomidine and ketamine is preferred by some pediatric sleep surgeons due to the lower risk of respiratory depression and upper airway obstruction as seen with other agents [5,17]. In this protocol, propofol is used which has a more rapid onset and shorter duration as compared to dexmedetomidine. Propofol can result in deeper sedation and more pronounced airway collapse while dexmedetomidine provides longer lasting sedation with little effect on upper airway obstruction [16-18-20]. During DISE, it is critical that the depth of sedation is closely monitored in coordination with the anesthesiologist. There is an ongoing debate regarding the best anesthetic protocol for DISE and more research with direct comparison of these agents is needed.

Conclusions

The future clinical value of pediatric DISE depends on the continuation of ongoing research and development of future studies. At this time large-scale institutional studies, systematic reviews, and meta-analyses which could improve the power of DISE research are limited by the lack of a universally accepted technique and scoring system. Here, we have proposed a specific protocol for scoring of DISE in children. More research is needed to determine the reliability of scoring systems used for DISE and to determine the optimal anesthetic protocol used during DISE.

Additional Information

Disclosures

Human subjects: Consent was obtained by all participants in this study. West Virginia University Office of Research Integrity and Compliance issued approval 17066146798001. Animal subjects: All authors have confirmed that this study did not involve animal subjects or tissue. Conflicts of interest: In compliance with the ICMJE uniform disclosure form, all authors declare the following: Payment/services info: All authors have declared that no financial support was received from any organization for the submitted work. Financial relationships: All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work. Other relationships: All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

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