

Predictors of Factors of Failure of DISE (Drug-Induced Sleep Endoscopy) Directed Adenoidectomy and/or Tonsillectomy in Children with Sleep-Disordered Breathing (SDB)

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Abstract

Background: Sleep-disordered breathing (SDB) is a common pediatric condition. Enlarged tonsils and adenoids are the commonest cause of SDB. However, many patients would have persistent signs and symptoms of SDB after Adenotonsillectomy (AT). Drug-induced sleep endoscopy (DISE) involves assessment of the upper airway using a flexible endoscope while patients are in a pharmacologically-induced sleep-like state. DISE may provide a more individualized surgical plan, predict factors that may cause treatment failure and limit unsuccessful surgeries.

Aim: Using DISE variables to predict the factors associated with persistent SDB in children after adenoidectomy and/or tonsillectomy. Patients and methods: This is a prospective non-randomized cross-sectional study included surgery-naïve patients (2-12 years of age) with adenotonsillar hypertrophy or infection and presented with symptoms of SDB. They have been referred to the Fayoum University Hospital for adenoidectomy and/or tonsillectomy. A pediatric sleep questionnaire (PSQ) for SDB was asked to be completed by the parents before and after the operation. Patients who underwent DISE-directed adenoidectomy and/or tonsillectomy for SDB between were eligible for the study. Other variables such as the history of asthma, Body mass index (BMI), snoring duration, history of allergic rhinitis, tonsil size according to Brodsky score, and adenoid size on X-ray were recorded. Children who had a previous operation for SDB and syndromic children are excluded. Results: One hundred patients have satisfied the inclusion criteria. Based on the postoperative modified PSQ, SDB resolved in 81 patients (81%), whereas 19(19%) had persistent symptoms. After DISE-directed adenoidectomy and/or tonsillectomy, allergic rhinitis (severe form), inferior turbinate hypertrophy, Deviated nasal septum (DNS), tongue base collapse, lateral pharyngeal wall collapse, lingual tonsil hypertrophy, and retroflexed epiglottis were associated with SDB treatment failure. Also, children who are obese and asthmatic were linked to unsuccessful surgery. Conclusions: DISE is a safe and useful technique for exact localization of sites of upper airway obstruction, improving surgical planning, elimination of unnecessary procedures, and improving surgical outcomes.

Keywords: Adenoidectomy; DISE; Pediatric sleep-disordered breathing; Tonsillectomy

Introduction

Pediatric Sleep-disordered breathing (SDB), a clinical spectrum ranging from simple snoring to Obstructive sleep apnea (OSA), is a highly prevalent condition that affects 4-11% of children [1,2].

Recurrent throat infections and chronic adenotonsillar hypertrophy associated with airway obstruction are the most common indications for Adenotonsillectomy (AT). AT is one of the most common pediatric surgical procedures performed [3]. AT was traditionally performed for recurrent tonsillitis and its sequelae but, in recent times, Sleep-disordered breathing (SDB)/Obstructive sleep apnea (OSA) in children has emerged as the primary indication for surgical removal of adenoids and tonsils [4].

Recent evidence has demonstrated that up to 30% of children undergoing AT for treatment of pediatric SDB and OSA will have a significant residual disease [2,4-6] likely due to obstruction at locations besides the tonsils or adenoids [7].

In order to decide a targeted and effective treatment plan, there is a growing consensus that an in-depth upper-airway evaluation is required to properly characterize the pattern of obstruction. For this aim, Drug-induced sleep endoscopy (DISE) is a method for evaluation of the upper airway using a flexible fiberoptic endoscope inserted transnasally during light sedation and spontaneous breathing that attempts to recreate natural sleep [8].

DISE allows direct observation of dynamic airway collapse throughout the upper airway. This technique has previously been shown to be reliable in both adults and children and has been shown to be able to discriminate snorers from non-snorers [9-11].

Non-responders to Adenotonsillectomy (AT) have mostly not been accounted for. Limited studies have suggested that variables such as obesity, asthma and severe Apnea-hypopnea index (AHI) are conferred risk for persistent disease after AT [2,12].

The aim of this study was to use DISE to evaluate both patient and DISE related findings as potential predictors of failure of adenoidectomy and/or tonsillectomy for SDB treatment in surgically naïve pediatric patients referred to the Fayoum University Hospital for adenoidectomy and/or tonsillectomy due to repeated infection or hypertrophy and with concurrent SDB.

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Patients and Methods

Patients

This is a prospective non-randomized cross-sectional study include patients selected from the ENT out-patient clinic of Fayoum University Hospitals between July 2017 & May 2018. Pediatric sleep questionnaire (PSQ) [13] was asked to be completed by the parents of every child who will undergo adenoidectomy and/or tonsillectomy before and after the operation.

Children who had abnormal (>33%) modified 22-item PSQ with adenoid and/or chronic adenotonsillitis and they are indicated adenoidectomy and/or adenotonsillectomy for that and aged between 2-12 years old were included in the study. Patients who had any craniofacial anomalies, dysmorphic features, or any associated genetic, neurologic, muscular, metabolic and any medical conditions or had any previous surgical management for SDB were excluded from the study.

History taking and clinical evaluation

Complete history taking including; history of asthma, prematurity at birth (<36 weeks gestation), snoring and its duration, persistent nasal discharge and symptoms of allergic rhinitis, and recurrent throat infections, and full ENT examination was done for all patients. Prior to surgery, tonsils size was graded according to the Brodsky score [14] and Body mass index (BMI) was assessed.

Lateral plain X-ray of the nasopharynx was done to evaluate the size of the adenoids and its relation to the size of the nasopharynx. The adenoid was considered small if it takes nearly 25% of the nasopharynx, moderate if takes nearly 50% and large if takes more than 75% of the nasopharynx [15].

Procedure

Parents gave written consent for adenoidectomy and/or tonsillectomy with DISE prior to the ongoing operation after a thorough explanation of the study. Before adenoidectomy and/or tonsillectomy, DISE was performed in the operating theatre. DISE was performed under sedation using total intravenous anesthesia of propofol &/ or midazolam which was given according to the bodyweight allowing spontaneous respiration [16].

Once a stable respiratory pattern was obtained, DISE was performed using a flexible fiberoptic endoscope. No local anesthesia or nasal decongestant was utilized. The nasal cavity was examined on both sides (the inferior turbinates, the nasal septum, and assessment of the condition of nasal mucosa). If accessible, the adenoid size was assessed in relation to the airway. Assessment of the retropalatal airway, the oropharynx, the tonsils, base of the tongue, vallecula, collapse of the pharyngeal airway an endolaryngeal airway was performed. Also, assessment of the epiglottis and the aryepiglottic folds and arytenoids were performed for any obstruction during respiration. Finally, the patient was intubated and mechanically ventilated then adenoidectomy and/or tonsillectomy were done.

Multi-level obstruction was defined as the presence of one or more upper airway abnormalities outside the adenotonsillar region.

A special scoring system that has been described in previous studies and has been shown to have good intra- and inter-rater reliability was adopted in this study with a total DISE score=14 and the least score=3 [16-18]. Arterial oxyhemoglobin saturation (pulse oximetry) was monitored throughout the procedure until complete recovery (Table 1).

Anatomic site	Description	Score
ERS [16]	No obstruction to either side, obstruction to one side, bilateral obstruction	1-3
NSD, inferior Turbinate hypertrophy & nasal polyp [16]	None, <50%, >50% obstruction of nasal airway patency	1-3
Adenoids (retropalatal vs. choanal)	<25%, 25-50%, 50-75%, >75%	1-4
Tonsils	<50%, >50%	0 or 1
Pharynx	No collapse, lateral wall, circumferential, anteroposterior, Tongue base collapse.	0 or 1
Lingual tonsil hypertrophy	No, Yes	0 or 1
Larynx	Normal, malacia, edema, Retroflexed epiglottis, folded epiglottis, polyp	0 or 1

ERS: Endoscopic Rhinitis Score; NSD: Nasal Septal Deviation

Table 1: Scoring system for Drug-induced sleep endoscopy (DISE).

All patients were asked to attend after 1 month of the operation to complete PSQ to assess the degree of improvement after the operation.

Statistical analysis of data

The organization, tabulation and statistically analysis of the collected data was performed by using Statistical product and service solutions (SPSS) software statistical computer package version 18 (SPSS Inc, USA). For quantitative data, the mean, standard deviation, median, and range were calculated. Independent t-test was used as a test of significance. For qualitative data, the number and percentage were calculated. As a test of significance, the Chi-square test or Fischer exact test was done when appropriate. For interpretation of the results of tests of significance, significance was adopted at $P \leq 0.05$.

Results

The study group consisted of 100 children, their age ranged from 2 to 12 years with mean age was (6.5 ± 2.6) years, fifty-four patients were males (54%) and forty-six patients were females (46%). The obesity rate was 10%. The prevalence of asthma among studied children was 12%. About 34% of children had history of allergic rhinitis (of whom 9 patients (26.5%) have mild symptoms, 13 patients (38.2%) have moderate symptoms and 12 patients (35.3%) have severe symptoms of allergic rhinitis while 66% (66 patients) have negative history of allergic rhinitis (Table 2).

According to Brodsky score for tonsil size grading, 2 children (2%) were grade 0, 12 children (12%) were grade 1, 32 children (32%) were grade 2, 38 children (38%) were grade 3 and 16 children (16%) were grade 4. Adenoid size according to the plain lateral X-ray of nasopharynx was less than 25% in 7 patients (7%), 26 patients (26%) have adenoid size about 50% % and 67 patients (67%) have adenoid size more than 75% (Table 2).

The mean of PSQ before the operation was (0.52 ± 0.15) as demonstrated in Table 2. This was reported as positive (> 0.33) that suggests a high risk for SDB. Post-operative the mean PSQ was (0.16 ± 0.12) , which indicated a postoperative overall improvement of SDB symptoms.

Total DISE score 14, each variable has its own score. Mean Endoscopic rhinitis score (ERS) score (1.98 ± 0.71) , Mean Deviated nasal septum (DNS) and inferior turbinate hypertrophy score (1.28 ± 0.60) , Mean Adenoid size score (3.35 ± 0.81) , Mean tonsil size score

Variable	Mean ± SD	Median (Range)
Age (years)	6.5 ± 2.6	6 (2-12)
Variable	N	%
Sex		
Male	54	54
Female	46	46
Obesity		
Obese	10	10
Normal weight	90	90
History of asthma		
Positive	12	12
Negative	88	88
Allergic rhinitis		
Positive	34	34
Mild	9	26.5
Moderate	13	38.2
Severe	12	35.3
Negative	66	66
Tonsil size (Brodsky score)		
Grade 0	2	2
Grade 1	12	12
Grade 2	32	32
Grade 3	38	38
Grade 4	16	16
Adenoid size according to X-ray		
25%	7	7
50%	26	26
≥ 75%	67	67
Type of operation		
Adenoidectomy (A)	16	16
Adenotonsillectomy (AT)	84	84
Pediatric sleep questionnaire (PSQ) score		
PSQ (pre-operative)	0.52 ± 0.15	0.50 (0.33-0.90)
PSQ (post-operative)	0.16 ± 0.12	0.11 (0.05-0.50)

Table 2: Demographic and clinical characteristics of the study population.

Variable	Mean ± SD	Median (Range)
ERS	1.98 ± 0.71	2 (1-3)
DNS and inferior turbinate hypertrophy	1.28 ± 0.60	1 (1-3)
Adenoid	3.35 ± 0.81	4 (1-4)
Tonsil	0.60 ± 0.49	1 (0-1)
Pharynx	0.12 ± 0.33	0 (0-1)
Larynx	0.01 ± 0.10	0 (0-1)
Tongue and lingual tonsil	0.15 ± 0.36	0 (0-1)
Total score	7.49 ± 1.99	7 (4-12)

ERS: Endoscopic Rhinitis Score; NSD: Nasal Septal Deviation

Table 3: DISE score of patients with SDB.

(0.60 ± 0.49), mean pharyngeal collapse score (0.12 ± 0.33), mean laryngeal view score (0.01 ± 0.10) and mean tongue base and lingual tonsil score (0.15 ± 0.36), Mean total DISE score (7.49 ± 1.99) (Table 3).

The mean BMI of patients (16.37 ± 1.76) who failed to have their symptoms resolved postoperatively was statistically significant (p-value <0.0001) compared to that of patients (15.07 ± 1.23) who succeeded to have their symptoms resolved postoperatively. It was demonstrated that the percentage of obese children (N=10) that succeeded to have their symptoms improved postoperatively (30%, 3 children) to that who have failed (70%, 7 children) which indicates that obesity is highly significant

Variable	Failed (N=19)		Succeeded (N=81)		p-value
	N	%	N	%	
Mean ± SD	6.1 ± 2.6		6.5 ± 2.7		0.537
Sex					
Male	11	20.4	43	79.6	0.705
Female	8	17.4	38	82.6	
Obesity					
Obese	7	70	3	30	<0.0001*
Normal weight	12	13.3	78	86.7	
History of asthma					
Positive	8	66.7	4	33.3	<0.0001*
Negative	11	12.5	77	87.5	
Allergic rhinitis					
Positive	16	47.1	18	52.9	<0.0001*
Negative	3	4.5	63	95.5	
Snoring duration					
Mean ± SD	3.3 ± 1.3		2.6 ± 1.3		0.034*
Tonsil size (Brodsky score)					
0	1 (50.0)	1 (50.0)	0	1 (50.0)	0.007*
1	1 (6.3)	11 (91.7)	1	1 (6.3)	
2	5 (15.6)	27 (84.4)	2	5 (15.6)	
3	4 (10.5)	34 (89.5)	3	4 (10.5)	
4	8 (50.0)	8 (50.0)	4	8 (50.0)	
Adenoid size					
25%	0	0	7	100	0.062
50%	2	7.7	24	92.3	
≥ 75%	17	25.4	50	64.6	
Operation					
Adenoidectomy	3	18.8	13	81.2	0.978
Adenotonsillectomy	16	19	68	81	

*Significant

Table 4: Relationship between failure of adenoidectomy or adenotonsillectomy to improve symptoms of SDB and demographic characteristics and patients' clinical and demographic characteristics.

predictor of persistence of symptoms and failure of operation (p-value .0001) (Table 4).

Twelve patients had a history of asthma, 8 of them (66.7%) showed treatment failure and 4 patients (33.3%) succeeded to have their symptoms resolved, that's indicated that history of asthma among children correlates significantly (p-value <0.0001) with success or failure of operation and persistence of symptoms. The total number of failed patients were 19, 16 of them have a positive history of allergic rhinitis and 3 patients have a negative history of allergic rhinitis. Positive history of allergic rhinitis was highly significant with persistence of symptoms of SDB postoperatively (p-value <0.0001), as shown in Table 4.

The average snoring duration in years in those who failed was (3.3 ± 1.3) in comparison to (2.6 ± 1.3) in those who responded to surgery (Table 4). It indicated that longer snoring duration is a statistically significant factor (p-value 0.034) for the persistence of symptoms after the operation. As shown in Table 4 the lower grade of tonsil size was significantly associated with improvement of symptoms after surgery.

The mean total DISE score for patient who failed {(19 patients out of 100 patients) was (10.42 ± 1.46)} and patients who succeeded {(81 patients out of 100 patients) was (6.80 ± 1.39)} to respond to operation was statistically significant (p-value 0.0001) (Table 5). According to ERS score DNS score and pharyngeal collapse score there was a significant

Variable	Failed (N=19)		Succeeded (N=81)		p-value
	N	%	N	%	
Allergic rhinitis & Inf. Turbinate hypertrophy					
Present	14	58.3	10	41.7	<0.0001*
Absent	5	6.6	71	93.4	
DNS & Inf. Turbinate hypertrophy					
Hypertrophy	2	66.7	1	33.3	0.033*
Normal	17	17.5	80	82.5	
Pharynx					
Lateral wall Collapse	8	66.7	4	33.3	<0.0001*
Normal	11	12.5	77	87.5	
Larynx					
Retroflexed Epiglottis	1	100	0	0	0.038*
Normal	18	18.2	81	81.8	
Tongue					
Lingual	8	80	2	20	<0.0001*
Normal	11	12.2	79	87.8	
Tongue base					
Enlarged base	5	83.3	1	16.7	<0.0001*
Normal	14	14.9	80	85.1	

*Significant

Table 5: Relationship between treatment failure and DISE score.

difference between those who successfully responded to surgery in comparison to those who failed (p values were <0.0001, 0.033 and <0.0001 respectively). Similarly, a significant difference in larynx and epiglottic score, lingual tonsil hypertrophy score and enlarged tongue base and its collapse score was observed with succeeded surgeries in comparison to failed surgeries (p values were 0.038, <0.0001 and <0.0001 respectively) (Table 5).

Discussion

DISE in surgically naïve patients had been reported only few in literature and has been reported to change the treatment decisions, in addition to enhancement of surgical achievements for SDB.

The aim of the current study was to look at the DISE variables that can predict failure of DISE-directed adenoidectomy and/or tonsillectomy in surgically naïve pediatric patients with SDB and to test the impact of DISE on surgery selection in these children.

The findings of this study have demonstrated that 19% (19 patients) failure rate in resolving SDB symptoms, based on modified PSQ, which was in accordance with the rates reported in other studies [2]. The importance of this study was the joint use of DISE-related variables and parent/patient reported outcome according to PSQ.

The disadvantage of this study is the unavailability of Polysomnogram (PSG) data. However, this study overcomes this defect by using PSQ. All patients included in this study scored positive on the modified PSQ (>.33) preoperatively, with different response to treatment postoperatively. Those who succeeded to have their symptoms resolved postoperatively had PSQ <.33, and those who failed and still complaining had their PSQ >.33.

Some investigators considered to make use of advanced radiological tools. Sleep cine Magnetic resonance imaging (MRI) allows full evaluation of the upper airway. In cases of adenoid and lingual tonsil hypertrophy, the depth of the tissue better appreciated on the MRI images. In addition, because there is no instrumentation of the airway, lesser amounts of anesthesia are needed for this procedure so the

evaluation may more closely represents natural sleep. Both static and dynamic images are obtained in both axial and sagittal views and can be reviewed multiple times. The main limitation is that this technique does not include an evaluation of the nasal airway, the larynx and the trachea. This technique is available on all MR scanners but is still new to many locations. Ideally, in cases of persistent OSA after AT, it would perhaps be best to get a sleep cine MRI first and then to consider a DISE procedure in the rare cases where the site(s) of obstruction are not identified by the sleep cine MRI. Advantages of DISE include the ability to see in 3 dimensions, as well as the ability to perform surgical correction concurrent with the DISE, thus limiting anesthesia exposure [19].

Boudewyns et al. [20] report was the first to demonstrate the findings of DISE and treatment outcomes of treatment in surgically naïve pediatric patients with SDB as our study subjects, i.e., with no syndromes or craniofacial abnormalities. The remaining pediatric studies which studied the DISE value to detect areas of upper airway obstruction in surgically naïve children were basically in special groups such as syndromic patients [17,21]. Other studies included both surgically naïve children suffering from SDB and those who had previous operations without exclusion of syndromic patients [18,22].

Results of this study demonstrated no statistical significance between demographic characteristics and response of the patient postoperatively. This is not consistent with other studies that stated that older male children (age >7) were more likely to show treatment failure [2,16].

It was found that longer snoring duration has an impact and correlates significantly with treatment failure. This may attribute to multiple airway level obstruction in these patients.

The demonstration that obese children were more likely to exhibit treatment failure that is in keeping with other studies. Tauman et al. [12] concluded that obese children were two times more possible to have an atypical post-operative Apnea/hypopnea index (AHI) than normal children.

Results showed that tongue base collapse and lingual tonsil hypertrophy were highly significant factors for treatment failure and demonstrated also the close relationship between tongue base collapse, obesity, and lateral pharyngeal wall collapse.

It was found that obese and overweight subjects were most likely to have tongue base obstruction and pharyngeal collapse on DISE. Our results showed that 10 children were obese, 9 of them had lateral pharyngeal wall collapse and 7 of them had tongue base collapse.

A recent meta-analysis [7] showed that AT significantly reduces the severity of OSA in obese children but is rarely curative with 75% to 88% of obese children with evidence of persistent OSA, which is close to our results (70% of obese children have persistent symptoms postoperatively).

Children who had a history of bronchial asthma were found to demonstrate a significant risk for persistence of symptoms postoperatively. Sleep-disordered breathing (SDB) and asthma are common and highly prevalent diseases in childhood. These two conditions share multiple common inflammatory pathophysiological pathways and are strongly connected by their position in the airway. As such, the possibility of existing interactions between asthma and SDB that affect their clinical course appears plausible, so proper treatment of asthma can greatly affect the resolution of symptoms of SDB postoperatively.

Boudewyns et al. [20] also reported that findings other than adenotonsillar obstruction were found in patients with SDB, such as palatal collapse, tongue base collapse, late-onset laryngomalacia which is also parallel to our study. However, they did not report any lingual tonsil hypertrophy nor did they demonstrate a relation between nasal pathology (such as significant chronic rhinitis, DNS or hypertrophied inferior turbinate) and treatment failure.

This study highlighted the impact of other factors in the nose that can be implicated in childhood SDB such as allergic rhinitis, inferior turbinate hypertrophy, and deviated nasal septum (especially severe forms).

Sullivan et al. [23] reported that nasal obstruction is rarely a primary cause of SDB, but it can worsen or cause apnea in children. Moreover, they demonstrated that persistent pediatric OSA is more common after AT in children who have severe persistent allergic rhinitis, enlarged inferior turbinate. The Radiofrequency (RF) treatment of inferior nasal turbinates is a suitable treatment in children with SDB that might be included in the treatment strategy for those children.

Large tonsils can be not obstructive during sleep if their pharyngeal muscles have a good tone with the maintenance of the oropharyngeal patency and if the tonsils tissue do not expand into the hypopharynx. While in other children with relaxed pharyngeal muscles during sleep even small tonsils may cause obstruction as the inferior pole that is usually hidden from view in the clinic, extends into the airway and causes obstruction.

This may help to interpret the results of Tang et al. [24] who studied children who had AT declaring that the tonsil size did not affect the apnea/hypopnea index, and that symptomatic recovery had been reported in non-obstructive tonsils according to the Brodsky scale. This drives future work to compare the size of tonsils by using both methods, and prevent unnecessary surgery based only on the routine trans-oral examination.

In this study, children with tonsil size grade 4 according to Brodsky score were 16 children, 8 of them (50%) showed significant improvement postoperatively. While on the other hand, 8 children (50%) expressed the persistence of symptoms and treatment failure. This can be explained on the basis of DISE findings before AT.

DISE in these patients showed that there were multiple anatomical sites other than adenoid and tonsil implicated in airway obstruction and causes persistence of SDB postoperatively.

In these patients with tonsil size grade 4 and showed treatment failure (8 children), other factors causing upper airway obstruction such as severe allergic rhinitis, marked inferior turbinate hypertrophy, obesity, lateral pharyngeal wall collapse, tongue base collapse, lingual tonsil hypertrophy, history of asthma, and retroflexed epiglottis.

This study is different and unique than other studies in the following:

- It uses PSQ as an evaluation tool both preoperatively and postoperatively so decreases the expenses of using PSG
- The factors of failure after AT were not just local anatomical obstruction in the upper airway but also systemic diseases as asthma, obesity and allergic rhinitis were also found to be contributed.
- There was not just one level of obstruction in the upper airway causing SDB but, with using DISE, multiple anatomical levels and systemic causes were incriminated.

- The tonsil and adenoid size were found not to be an indication of the severity of SDB and improvement post AT.
- The study was undertaken in surgically naïve children with history of SDB. Children who had a previous operation for SDB and syndromic children were excluded.

Conclusion

DISE is a safe and useful technique for exact localization of sites of upper airway obstruction, improving surgical planning in children with SDB, elimination of needless procedures, and enhancement of surgical outcomes. Other factors attributed for failure after AT according to DISE findings in this study were mainly children who had allergic rhinitis, marked inferior turbinate hypertrophy, deviated nasal septum (severe forms), lateral wall pharyngeal collapse, tongue base collapse, lingual tonsil hypertrophy, and occult laryngomalacia. It was found, also that obesity and asthma are important predictors of treatment failure.

Conflict of Interest

None

Ethical considerations

The study was approved by the Ethical Committee, Faculty of Medicine, Fayoum University and followed the good clinical practice guidelines of the Helsinki's World Medical Association Declaration.

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