**Drug induced sleep endoscopy for targeted sleep surgery in pediatric patients**

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**Abstract**

**Introduction**

Patients with obstructive sleep apnea (OSA) and sleep disordered breathing are commonly managed by surgical intervention targeting the anatomic obstruction. Untreated OSA can lead to detrimental effects in children including daytime sleepiness and poor cognition1. Adenotonsillectomy has been shown to effectively improve the apnea hypopnea index (AHI), and sleep related quality of life, and the current recommendations suggest removal of tonsils and adenoids as first line treatment of pediatric OSA2. However, not all patients with symptoms of sleep disordered breathing or positive sleep studies present with classical physical exam finding of adenotonsillar hypertrophy. These patients are more clinically challenging. Drug induced sleep endoscopy (DISE) has become an increasingly popular technique to evaluate the airway and the level of obstruction in children with clinically small tonsils and adenoids3. Past studies using DISE have demonstrated that clinically small tonsils (size 1 to 2+) may not be a cause of obstruction4 and have questionedadenotonsillectomy as the first line treatment.

DISE has been shown to be an effective tool in optimizing surgical intervention for pediatric OSA. A recent study by He et al. evaluated preoperative and post-operative AHI in children with history of adenotonsillectomy undergoing DISE directed surgery and found a significant decrease in AHI following surgery5.

This study aims to determine the anatomical level of obstruction in children undergoing DISE, whether there is a correlation between location of obstruction and/or surgery performed on outcomes and the efficacy of DISE directed surgery for OSA.

**Methods**

This was a prospective cohort study performed on patients who underwent DISE from January 2017- October 2017 by one of two fellowship trained pediatric otolaryngologists. All study protocols were approved by the Beaumont Health Institutional Review Board. Subjects completed validated pre-operative University of Michigan Pediatric Sleep Questionnaire (UMPSQ)6.

Inclusion criteria were patients ages 6 months to 18 years with the diagnosis of OSA or sleep disordered breathing. Exclusion criteria was pregnancy and patients with size 4 tonsils. All children were scheduled for DISE prior to surgical intervention for sleep disordered breathing and OSA. OSA severity was defined based on AHI: mild was 1 to <5 events per hour, moderate was 5 to <10 and severe as >10.

*Validated Pediatric Sleep Questionnaire*

Pre and post-operative University of Michigan Pediatric Sleep Questionnaire (UMPSQ) were given to determine the likelihood of residual OSA. Prior to surgery, parents or guardians of subject participants completed a 22 question validated pediatric sleep questionnaire. The total number of yes responses were calculated to give a pre-operative score. Parents were asked to complete the survey again at least 4 weeks post operatively. Post-operative surveys were either filled out at the post-operative office visit or were completed via telephone.

*DISE Directed Surgery*

Drug induced sleep endoscopy was performed at Beaumont Children’s Hospital or at outpatient surgery center. DISE technique was performed as described by Wooten7. Once in the operative suite, patients are masked under sevoflurane/nitrous oxide inhalation anesthetic. Intravenous access is then achieved and propofol is delivered in 1 mg/kg boluses the by the department of anesthesia to achieve a level of consciousness to mimic natural sleep (asleep but spontaneously breathing). A flexible fiberoptic endoscope was then passed through the nose and the airway was assessed. Findings recorded during DISE included tonsil size, adenoid size, Yellon Tongue base score (8), lateral pharyngeal wall (LPW) collapse and evidence of laryngomalacia. Adenoids were graded in percentage obstruction (0-100%). Tonsils were graded on percentage of obstruction (0-100%). Lateral pharyngeal wall collapse was determined to be present or absent. Yellon Tongue base score was calculated as described by Yellon; Grade 0 is a normal airway, Grade 1 is prolapse of the epiglottis along posterior pharyngeal wall but normal position of the tongue, Grade 2 is prolapse of the epiglottis and base of tongue with only epiglottic tip visible, Grade 3 is glossoptosis with no portion of the epiglottis visible8. Laryngomalacia was assessed as present or absent. Surgery was performed as per the surgeon’s clinical judgment based on location of obstruction. Surgeries performed included: tonsillectomy, adenoidectomy, lingual tonsillectomy, lateral pharyngoplasty and supraglottoplasty.

*Statistical analysis*

Patient charts were reviewed for patient demographics including age and sex, pre-operative apnea hypopnea index, pre-and post-operative sleep quality of life scores. Findings on DISE as described above and surgical procedure performed were recorded. Statistical analysis was performed using The SAS System for Windows version 9.4. Frequency and percentages were determined for DISE findings including tonsil size, adenoid size, Yellon tongue base score, presence of laryngomalacia and lateral pharyngeal wall collapse. Lateral pharyngeal wall collapse present was compared to tonsil size and percentages of collapse present were determined for 1+, 2+, and 3+ tonsils (as well as a collapsed version of small versus large). Patients with 4+ tonsils were not included because these patients did not need DISE and typically underwent tonsillectomy.

For variables with a normal distribution mean and standard deviation were reported and group comparisons used a two-sample t test and a 95% confidence interval for the difference in means. For continuous variables without a normal distribution the median was calculated and p-values for group comparisons were obtained using a Wilcoxon Rank-Sum test. Comparison of whether the surgery performed was the recommended adenotonsillectomy with LPW, AHI and tonsil size was done using an exact version of Pearson’s chi-square test and an exact 95% confidence interval for the odds ratio.

**Results**

There were 42 patients who met the inclusion criteria. Of the study participants there were 10 females (23.8%) and 32 males (76.2%). Ages ranged from 1-17 years and the median age was seven years old. 32 patients had pre-operative polysomnograms (PSGs) and of these patients 16 had mild OSA (38.1%), 12 moderate (28.6%) and 14 severe OSA (33.3%). The median apnea hypopnea index (AHI) was 6.8 (range 1  – 106) (table 1).

Pre-operative DISE findings were recorded and 9 patients (21.4%) had 1+ tonsils, 24 patients (57.1%) had 2+ tonsils, 9 patients (21.4%) had 3+ tonsils. Yellon Tongue Base Score was 0 in 11 patients (26.2%), 1 in 11 patients (26.2%), 2 in 12 patients (28.6%) and 3 in 8 patients (19.1%). Laryngomalacia was identified on endoscopy in 7 patients (16.7%). Lateral pharyngeal wall collapse was noted in 21 patients (50%) (table 2). Surgeries were performed based on anatomic obstruction shown with DISE and are summarized in table 3.

LPW collapse was associated with increasing tonsil size. Of patients with size 1 tonsils 22.2% had LPW collapse, 41.7% of patients with size 2 tonsils had LPW collapse and 100% of patients with size 3 tonsils had LPW collapse. There was a statistically significant positive correlation between tonsil size and LPW collapse (P = 0.0015). There was a slightly inverse correlation between tonsil size and OSA severity. Patients with smaller tonsils were more likely to have severe OSA, 66.7% of size 1 tonsils had severe OSA while only 22.2% of patients with 3+ tonsils had severe OSA (table 4).

More than half of the patients did not have the academy guidelines recommended adenotonsillectomy (n = 27, 64.3%). Patients with larger tonsils were more likely to have adenotonsillectomy, 89% of patients with 3+ tonsils had a T&A, 25% with 2+ tonsils had a T&A and only 11% with 1+ tonsils had T&A (P = 0.001). However, patients with higher Yellon tongue base score were less likely to have T&A, 12.5% of patients with Yellon tongue base 3 had a T&A compared to 64% of patients with Yellon 0 (P=0.005). There was insufficient information to draw a conclusion about the existence of a relationship between OSA severity and whether adenotonsillectomy was performed (P = 0.42) (table 5).

*UMPSQ Findings*

Twenty-five patients (60%) completed the post-operative survey. The remainder of patients could not be reached. Overall, patients had improved in UMPSQ scores from 13.4 ± 3.7 prior to surgery to 5.7 ± 3.5 after surgery.  The average change from pre-operative score to post-operative score was 7.7 ± 4.5. Patients who underwent adenotonsillectomy had a greater decrease in scores than those who did not, the average change for patients who had adenotonsillectomy was 9.8 ± 4.5, while the average change for patients who did not was 6 ± 3.9 (P = 0.032). There was no clear relationship between tonsil size and amount of improvement of survey results. Similarly, there was no well-defined relationship between pre-operative LPW collapse or OSA severity and change in UMPSQ scores (table 6).

Patients who filled out the postop survey had slightly higher average pre-operative survey scores (13.4 vs 11.9), and were more likely to have tonsillectomy (72% of patients who only underwent tonsillectomy completed a post-op survey, while 43% of patients who had other surgeries completed the post-operative survey). Patients who had severe OSA were also more likely to complete the post-op survey (completion: 79% for severe, 50% for mild and moderate). Patients with LPW were more likely to complete the post-op survey, 71% with LPW versus 48% without LPW collapse. Postop surveys were available for 44%, 58%, 78% of patients with tonsil sizes 1, 2, 3 respectively.

**Discussion**

This study presents the use of DISE in the treatment of pediatric OSA or sleep disordered breathing as a tool to determine the anatomical level of obstruction and explores the correlation between this, OSA severity and outcomes. A validated sleep survey administered pre-operatively and post operatively determined the efficacy of surgery. Pre- and post-operative PSQ sleep questionnaires showed that surgery was effective in the treatment of OSA and sleep disordered breathing and has positive impact on quality of life. Average post-operative score was significantly less than pre-operative score (mean decrease of 7.7 ±4.5, p=0.05). Prior studies have similarly demonstrated efficacy in DISE directed surgery measured by a significant decrease in AHI5.

The majority of patients who underwent DISE had clinically small tonsils, 79.6% of patients had 1+ or 2+ tonsils. This is valuable to note as children with history consistent with OSA and/or a positive sleep study often have large adenoids that are may not be visualized on physical exam unless fiberoptic scope is performed. However, clinically small tonsils should not be by default considered non-obstructive. LPW collapse was noted in 22.2% of patients with 1+ tonsils and 41.7% of patients with 2+ tonsils, which are usually considered clinically small. Prior studies have evaluated the significance of clinically small tonsils on airway obstruction using DISE. In a retrospective review Miller et al reported a correlation between tonsil size and lateral pharyngeal wall collapse4. In their results they stated that of patients with a Brodsky score of 1, 60% had a LPW score of 0. However, they did not report on the frequency of obstruction in patients with 2+ tonsils. These finding are in congruence with this study, tonsil size positive correlates with LPW obstruction.

A variety of surgical procedures were done based on intra-operative DISE findings, therefore, surgery was categorized as recommended versus not recommended, based on the current academy guidelines for treatment of pediatric OSA. Recommended surgery was performed less often than not recommended surgery and 64.3% of children had findings on DISE that necessitated more complex surgery than adenotonsillectomy such as supraglottoplasty, lingual tonsillectomy, laryngeal cleft surgery, turbinate reduction or a combination of these procedures. A 2017 study by Gazzaz suggested that DISE findings led to deviation from recommended adenotonsillectomy in 35% of patients9. Similarly to this study, their classification adenoidectomy alone or tonsillectomy alone was considered deviation from standard of care.

The most common intervention performed outside of adenotonsillectomy was lingual tonsillectomy (38.1%). This differs from the study by Miller et al where the most common alternative intervention was supraglottoplasty, which was done in 43% of patients4. In this study, only 11.9% of patients underwent supraglottoplasty.

When comparing whether the recommended surgery was done for varying levels of OSA severity, the data revealed that AHI may lower in patients undergoing adenotonsillectomy. However, this was not found to be statistically significant (p = 0.42). As expected, larger tonsil size did correlate with increased likelihood for adenotonsillectomy. Meanwhile, higher yellon tongue base scores correlated with less likelihood of having the recommended adenotonsillectomy (p = 0.005). This corroborates with previous research suggesting that children with clinically small tonsils likely have obstruction at other sites4,10.

Interestingly, patients who underwent the recommended adenotonsillectomy had significantly higher change from pre-operative to post-operative scores than patients who underwent other surgery. This may indicate that obstruction at the level of the adenoids and tonsils is easier to treat than obstruction further down in the airway. However, more patients who had adenotonsillectomy completed the post-operative survey, 72% of those who underwent adenotonsillectomy versus only 43% of patients who had other surgeries completed the post-operative survey. Patients who had severe OSA were also more likely to complete the post-operative survey (completion: 80% for severe, 50% for mild and moderate). Further research with a larger sample size is warranted to address this.

There are several limitations of this study. Not all patients had a pre-operative PSG and no post-operative PSGs were done. In future studies, this would be a valuable objective measure to analyze. Only 25 of 42 patients completed the post-operative UMPSQ and with a larger sample size, more statistically significant correlations may be seen. Additionally, there was no control group.

**Conclusion**

This study demonstrates thatDISE is a valuable tool for evaluation of multi-level obstruction in pediatric patients with OSA or sleep disordered breathing. Similar to findings of previous studies, DISE often changes surgical management from traditional adenotonsillectomy to surgery to address the anatomic level of obstruction. This is increasingly important for patients with small adenoids and tonsils because the level obstruction lies further in the airway and can’t be seen on physical exam.

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Table 1. Patient demographics and OSA severity.

|  |  |  |
| --- | --- | --- |
| **Age, yr.** (median, range) | 7  | 1-17 |
| **Gender** | **N** | **%** |
| F | 10 | 23.8% |
| M | 32 | 76.2% |
| **OSA severity** | **N** | **%** |
| mild | 16 | 38.1% |
| moderate | 12 | 28.6% |
| severe | 14 | 33.3% |
| **AHI** (median, range) | 6.8  | 1– 106 |

Table 2. Preoperative drug induced sleep endoscopy (DISE) findings.

|  |  |  |
| --- | --- | --- |
| **Finding** | **N** | **%** |
| **Adenoid Obstruction**  |   |  |
| yes | 29 | 31.0 |
| no | 13 | 69.1 |
| **Tonsil Size**  |  |  |
| 1+ Tonsil | 9 | 21.4 |
| 2+ Tonsil | 24 | 57.1 |
| 3+ Tonsil  | 9 | 21.4 |
| **Yellon Tongue Base**  |  |  |
| 0 | 11 | 26.2 |
| 1 | 11 | 26.2 |
| 2 | 12 | 28.6 |
| 3 | 8 | 19.1 |
| **LPW Collapse**  |  |  |
| no | 21 | 50 |
| yes | 21 | 50 |
| **Laryngomalacia**  |  |  |
| no | 35 | 83.3 |
| yes | 7 | 16.7  |

Table 3. Surgeries performed.

|  |  |  |
| --- | --- | --- |
| **Surgery Performed** | **N** | **%** |
| Tonsillectomy | 23 | 54.76% |
| Adenoidectomy | 35 | 83.33% |
| Lingual Tonsillectomy | 16 | 38.10% |
| Supraglottoplasty | 5 | 11.90% |
| Laryngeal cleft | 2 | 4.76% |
| Turbinate reduction | 2 | 4.76% |

Table 4. Relationship of tonsil size with LPW collapse and OSA severity. Percentages indicate how many patients with given tonsil size had LPW collapse and different severities of OSA.

|  |  |  |  |  |
| --- | --- | --- | --- | --- |
| **Tonsil size** | **LPW collapse** | **Mild OSA** | **Moderate OSA** | **Severe OSA** |
| 1 (n = 9) | 222.2% | 222.2% | 111.1% | 666.7% |
| 2 (n = 24) | 1041.7% | 1041.7% | 833.3% | 625% |
| 3 (n = 9) | 9100.0% | 444.4% | 333.3% | 222.2% |

Table 5. Relationship between tonsil size, yellon tongue base score, OSA severity and whether academy guideline recommended adenotonsillectomy was performed. \*Percentages indicate the number of patients who had the variables listed in the columns.

|  |  |  |  |  |
| --- | --- | --- | --- | --- |
|  |  | **Tonsil Size** | **Yellon score** | **OSA Severity** |
| **T&A**  | Total N%\* | **1+** **n = 9** | **2+** **n = 24** | **3+****n = 9**  | **0****n = 11** | **1****n = 11** | **2****n = 12** | **3****n = 8** | **Mild****n = 16** | **Mod** **n = 12** | **Severe****n = 14** |
| No  | 27 64.3% | 8 88.9% | 18 75% | 1 11.1% | 4 36.4% | 6 54.6% | 10 83.3% | 7 87.5% | 9 56.3% | 8 66.7% | 10 71.4% |
| Yes  | 1535.7%  | 1 11.1% | 6   25%  | 8 88.9% | 7 63.6% | 5 45.5% | 2 16.7% | 1 12.5% | 7 43.8% | 4 33.3% | 4 28.6% |
|   |   | P = 0.001 | P = 0.005 | P = 0.42 |

Table 6. Mean change from pre-operative to post-operative UMPSQ surveys based on surgery performed, OSA severity, tonsil size and LPW status.

|  |  |  |  |
| --- | --- | --- | --- |
| **Group** | **N** | **Mean change in survey score** | **SD** |
| All patients | 25 | 7.68 |

|  |
| --- |
| 4.49 |

 |
| Surgery performed |
| Not T&A | 14 | 6.0 |

|  |
| --- |
| 3.88 |

 |
| T&A | 11 | 9.82 |

|  |
| --- |
| 4.45 |

 |
|   |   |   | P = 0.032 |
| OSA severity |
| mild | 8 | 10 | 4.5 |
| moderate | 6 | 6.33 | 3.08 |
| severe | 11 | 6.73 | 4.80 |
|   |   |   | P = 0.21 |
| Tonsil size |
| 1 | 4 | 7.75 | 5.19 |
| 2 | 14 | 7.29 | 4.89 |
| 3 | 7 | 8.43 | 3.78 |
|   |   |   | P = 0.87 |
| LPW collapse |
| No | 10 | 6.20 | 3.88 |
| Yes | 15 | 8.67 | 4.72 |
|   |   |   | P = 0.18 |