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ORIGINAL RESEARCH

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Drug-induced sleep endoscopy effect on intraoperative decision making in pediatric sleep surgery: A 2-year follow up

Robert M. Frederick II MD | Josiah Brandt BS | Anthony Sheyn MD

Department of Otolaryngology, College of Medicine, University of Tennessee Health Science Center, Memphis, Tennessee, USA

Correspondence
Robert M. Frederick II, Department of Otolaryngology, College of Medicine, University of Tennessee Health Science Center, 910 Madison Avenue Ste 430, Memphis, TN 38163, USA.
Email: robertfrederick2@gmail.com

Abstract

Objective: To further demonstrate sleep endoscopy's utility in improving surgical outcomes for pediatric OSA.

Methods: This is a retrospective review of surgically naïve patients <18 years old with diagnosed moderate–severe OSA who underwent DISE at the time of initial sleep surgery. Patients included in final analysis had both preoperative and postoperative polysomnograms. Surgical success was defined as an oAHI decrease by at least one diagnostic category. Residual OSA was defined as any patient with postoperative oAHI >1.

Results: A total of 106 patients had preoperative and postoperative polysomnograms. Patients with comorbidities comprised 53.8% of the group. Average BMI% was 88.2, with 75.5% classified as obese. The most common area of collapse was the base of tongue, occurring in 32.1% of patients. There was a statistically significant decrease from the mean preoperative oAHI of 29.7 to the mean postoperative oAHI of 6.6 ($p < 0.001$). Surgical success occurred in 76.4% of patients. A postoperative oAHI of <5 was achieved in 57.7% of patients with moderate or severe OSA. The average BMI% of patients who met surgical success was 86.4, while the average BMI % of patients who did not was 90.8. A postoperative oAHI of <5 was achieved in 68.4% of patients with a BMI% < 85, compared with 55.2% of patients with a BMI% ≥ 85.

Conclusion: This study supports the utilization of DISE during initial surgery for severe sleep apnea in the pediatric population. It was found to effectively aid in significantly reducing surgically naïve patients' mean oAHI.

Level of Evidence: Level III.

KEYWORDS
OSA, pediatric, residual OSA, sleep endoscopy, sleep surgery

1 | INTRODUCTION

Obstructive sleep apnea (OSA) is a common pediatric sleep disorder with a prevalence of up to 6%.[1,2] Repeated episodes of complete or partial airway obstruction leads to sleep fragmentation and hypoxia in patients with this disorder. OSA is frequently associated with adenotonsillar hypertrophy in pediatric patients.[1] Other causes of OSA include enlargement of the base of tongue, uvula, soft palate, or lingual tonsils, as

well as nasoseptal obstruction, micrognathia, maxillary hypoplasia, and pharyngeal hypotonia.[3]

In the setting of confirmed adenotonsillar hypertrophy, first-line treatment for OSA involves adenotonsillectomy (AT).[3] It has been shown that between 15% and 20% of children are found to have persistent OSA refractory to primary AT.[2,4,5] This rate of persistent OSA is likely due to obesity and other factors which cause increased upper airway obstruction.[5–7] Surgical treatments for persistent OSA unresolved after AT include lingual tonsillectomy, septoplasty, supraglottoplasty, base of tongue resection, pharyngoplasty, and uvulopalato-pharynoplasty.[6] Drug-induced sleep endoscopy (DISE) is useful for identifying sites of obstruction prior to surgery by providing direct visualization of the upper airway during anesthesia.[8]

DISE has been demonstrated to safely and effectively aid in determining the location and degree of upper airway obstruction, with moderate to substantial interrater reliability and good test re-test reliability.[5,9] Specific DISE findings include the severity of palatal and hypopharyngeal obstruction, the degree of epiglottis, tongue, velum, and lateral wall collapse, and the effects of mouth opening on airway obstruction.[7] DISE has also been shown to effectively influence the choice of therapy in adults with OSA, potentially improving surgical outcomes in initial OSA treatment.[8,10,11] Site-specific surgical treatment for patients with complex OSA can be guided by DISE findings, one study showed that surgical decision making changed in 50.24% of cases following DISE.[12] For these reasons, DISE may be used as a standard procedure for adults with severe OSA.[11]

Historically, DISE was used for pediatric patients who did not respond to initial AT, as an attempt to treat their persistent OSA.[4,7] More recently, DISE utilization has expanded for surgically naïve pediatric patients in specific scenarios, such as patients at high risk for persistent OSA (Down syndrome, obesity, neurologic disorders, etc.), patients with severe OSA yet small tonsils and adenoids, patients with known sleep-state dependent laryngomalacia; or patients being evaluated as potential candidates for a hypoglossal nerve stimulator procedure.[13–16]

The purpose of this study is to expand upon the findings of a previous study conducted by the senior author that demonstrated sleep endoscopy's utility in improving surgical outcomes for pediatric OSA. The prior study showed that utilizing DISE led to more significant improvements in patients' oAHI and the possibility of completely resolving some patients' OSA. With a larger cohort of patients and additional statistical support, the objective of this study is to build upon the previous study's results and solidify the role of DISE in intraoperative decision making for pediatric OSA.

2 | MATERIALS AND METHODS

Approval was granted from the University of Tennessee Health Science Center IRB. A retrospective chart review was performed on patients at Le Bonheur Children's Hospital. Subjects were surgically naïve, under 18 years old, and met criteria for moderate–severe OSA based on preoperative PSG with an obstructive apnea-hypopnea index (oAHI) ≥ 5. Following standard practice, informed consent from caregivers of patients undergoing DISE was obtained to include all possible sleep surgery procedures deemed necessary. In the process of obtaining consent, a discussion was had with all caretakers about the complications and risks of each procedure that would possibly be performed during surgery depending on the DISE findings. These included DISE, tonsillectomy, adenoidectomy, pharyngoplasty, base of tongue reduction, lingual tonsillectomy, turbinate reduction, and supraglottoplasty. Variables collected from the electronic medical record included age, gender, race, location of obstruction, type of surgical intervention, and pre- and postoperative oAHI. Reported physical exam findings, if present, include tonsillar and turbinate hypertrophy. The duration of DISE was not measured specifically; duration of every DISE procedure was included in the total surgical time. Findings of DISE were classified at the level of airway obstruction. Due to universal adenotonsillar hypertrophy findings, each patient underwent adenotonsillectomy following DISE. Additional surgeries performed after DISE and concurrently with AT included inferior turbinate reduction, supraglottoplasty, expansion pharyngoplasty, and lingual tonsillectomy with midline base of tongue reduction A paired-samples *t*-test was used to compare preoperative and postoperative oAHI in patients who had postoperative PSG follow-up.

3 | RESULTS

A total of 213 patients were identified with documented DISE findings that met the inclusion criteria. 106 of the total 213 patients had both preoperative and postoperative polysomnograms (PSGs) and were thus included in the analysis. Ages ranged from 1 to 18 years old with a mean age of 8.4 (SD 4.57). The gender distribution included 34 (32%) females and 72 (68%) males. Racial/ethnic backgrounds were reported as 75 (70.8%) African American, 21 (19.8%) White, 7 (6.6%) Hispanic, 2 (1.8%) Asian, and 1 (0.9%) other. A total of 57 (53.8%) patients had medical comorbidities. 19 patients had heart conditions (structural abnormality, arrhythmia), 16 had endocrine abnormalities (acanthosis nigricans, hyper-insulinemia, Type 2 Diabetes Mellitus), 18 had non-anatomic upper airway disorders (i.e., asthma, allergic rhinitis), and 15 had a developmental or speech delay. Average body mass index percentile (SD) was 88.2 (27.4). A total of 80 (75.5%) patients were classified as obese, corresponding to a BMI% ≥ 95; whereas 7 (6.6%) patients were classified as overweight, corresponding to a body mass index percentile (BMI%) between 85 and 95.

Although specific tonsil size was not recorded, adenotonsillar hypertrophy was a universal physical exam finding for all of the patients, thus each patient underwent AT with or without concurrent procedures. Other common sites of obstruction found on DISE were recorded as well. Of the 106 patients whose DISE findings were reviewed, the most common area of collapse was the base of the tongue, occurring in 34 (32.1%) patients. Soft palate collapse was found in 23 (21.7%) patients. Obstruction involving the epiglottis was found in 14 (13.2%) patients. Evidence of multilevel collapse was observed in 11 (10.4%) patients (Table 1). A total of 29 (27.4%) patients observed to have palatal collapse during DISE underwent a pharyngoplasty at the time of AT. Table 2 outlines other

TABLE 1 DISE findings

Site of collapse	Number (%)
Tonsils and adenoids	106 (100)
Base of tongue	34 (32.1)
Soft palate	23 (21.7)
Epiglottis	14 (13.2)
Multilevel collapse	11 (10.4)

TABLE 2 Concurrent procedures

Procedure performed with adenotonsillectomy	Number (%)
Pharyngoplasty	29 (27.4)
Tongue reductions	22 (20.1)
Inferior turbinate reduction	13 (12.2)
Supraglottoplasty	6 (5.7)

procedures performed during AT, directed by DISE findings, included 22 (20.1%) tongue reductions, 13 (12.2%) inferior turbinate reductions, and 6 (5.7%) patients who underwent a supraglottoplasty.

The mean preoperative oAHI (SD) was 29.7 (25). The mean postoperative oAHI (SD) was 6.6 (6). The mean length of time between surgery and postoperative PSG (SD) was 4.2 months (2.8). A paired-samples *t*-test showed a statistically significant decrease between mean pre- and postoperative oAHI ($p < 0.001$), indicating improvement of oAHI after DISE-assisted surgical intervention (Table 3). The overall surgical success rate, defined as an oAHI decrease by at least one diagnostic category (i.e., severe OSA (oAHI >10) reduced to moderate OSA (oAHI 5–10) or better, moderate OSA reduced to mild OSA (oAHI 1–5) or better, or mild OSA reduced to no OSA (oAHI ≤1)), was found to be 81/106 (76.4%). Postoperatively, 22 patients (20.8%) had severe OSA, 23 (21.7%) had moderate OSA, 53 (50%) had mild OSA, and 9 (8.5%) had no residual OSA. Among the 104 patients with severe or moderate preoperative OSA, 60 (57.7%) patients had mild or better postoperative OSA (oAHI <5), which is commonly used as a diagnostic value for recommending CPAP utilization.

Residual OSA, any patient with postoperative oAHI >1, occurred in 97 (91.5%) patients. The average preoperative oAHI was 30.4 in the patients with residual OSA, and 22.3 in the patients with no residual OSA. African American patients had residual OSA 93.3% of the time, while 80.9% of white patients had residual OSA. Of the 57 patients with medical comorbidities, 51 (89.5%) had residual OSA, while 46 of the 49 patients with no medical comorbidities (93.6%) had residual OSA. In the nine patients with no residual OSA, 6 (66.7%) patients had medical comorbidities. In the 97 patients with residual OSA, 46 (47.4%) patients had medical comorbidities. In total, 81/87 (95.4%) patients with BMI % ≥ 85 experienced residual OSA; compared with 16/19 (84.2%) patients with BMI% < 85 who experienced residual OSA (Table 4). The average BMI% of patients with residual OSA was 88.2. The average BMI% of patients with no residual OSA was 78.9 ($p = 0.34$).

Among patients with BMI% ≥ 85, 61/87 (76.3%) met surgical success criteria; compared with 20/26 (76.9%) patients with BMI% < 85. The average BMI% of patients who met surgical success criteria was 86.4. The average BMI% of patients who did not meet surgical success criteria was 90.8. The number of patients with a postoperative oAHI <5 was 48/87 (55.2%) for patients with BMI% ≥ 85; compared with 13/19 (68.4%) for patients with BMI% < 85.

Among the 57 patients with medical comorbidities, 40 (70.2%) patients met surgical success criteria, 18 (31.6%) patients had an 85% reduction in oAHI, and 28 (49%) had a postoperative oAHI <5. Among the 49 patients without medical comorbidities, 41 (83.7%) patients met surgical success criteria, 24 (48.9%) patients had an 85% reduction in oAHI, and 32 (65.3%) had a postoperative oAHI <5. Average percentages of oAHI reduction grouped according to medical comorbidity were as follows: airway (76.7%), endocrine (66.9%), cardiac (47.6%), and developmental/speech (37.7%). Patients with no listed medical comorbidities had an average oAHI reduction of 75.7%. No adverse events related to sleep endoscopy or the ensuing multi-level airway surgeries were reported. Specifically, no complications arose from the extended anesthesia time required to perform sleep endoscopy.

4 | DISCUSSION

There are many known risk factors for pediatric OSA such as obesity, Down syndrome, neurologic disorders, achondroplasia, among others.[1] Higher rates of OSA and more severe OSA, determined by a higher oAHI measurement, are also more common in certain races and ethnicities, with non-Hispanic African American and Hispanic children affected more often than others.[1] OSA negatively impacts the child's quality of life. Those experiencing the disease do not achieve adequate or restful sleep. Instead, they endure snoring, enuresis, and apneic episodes. This leads to daytime sleepiness, inattention, irritability, hyperactivity, and subsequent poor academic performance and a decline of memory and cognitive performance.[1–3,17] Consistent with the literature on signs and symptoms of pediatric OSA, the patients in this study experienced snoring, daytime sleepiness, and developmental or speech delays.

The American Academy of Pediatrics (AAP) recommends adenotonsillectomy (AT) as a first line treatment for pediatric OSA.[18] However, not every child will experience resolution of symptoms from this surgery alone, and 54–76% of children will go on to experience residual OSA.[18,19] The rate of residual OSA depends on the definition used. Our study determined residual OSA to be marked by a postoperative oAHI >1, a common cutoff point in the literature.[20] Therefore, patients were classified as having residual OSA even if they experienced a dramatic reduction in oAHI between preoperative and postoperative measurements or if their persistent objective findings qualify as mild disease. Postoperative AHI <2 and <5 have also been used to define residual OSA.[19,21,22] This small difference in objective measurement for residual OSA definition leads to large differences in the rates of reported residual OSA, with one study demonstrating a range between 20% and 72%.[23] Regardless of definition, there are known risk factors for experiencing residual OSA following AT, many of which overlap with the risk factors for developing pediatric OSA.

TABLE 3 Comparison of preoperative and postoperative obstructive apnea hypopnea index

Group	Preoperative mean (SD, range)	Postoperative mean (SD, range)	*p* value
Entire cohort, *N* = 106	29.7 (25.1, 5.5–178.9)	6.6 (6, 0.4–34)	$p < 0.001$
Gender			
Male, *N* = 72	29.1 (26.5, 5.5–178.9)	7.3 (6.5, 0.5–34)	$p < 0.001$
Female, *N* = 34	30.9 (21.5, 6–98.4)	4.9 (4.4, 0.4–19.6)	$p < 0.001$
Race			
African American, *N* = 75	30.9 (26.9, 6.3–178.9)	3.9 (6.2, 0.4–34)	$p < 0.001$
White, *N* = 21	22.2 (5.6, 5.5–96.5)	5.6 (4, 0.7–12.8)	$p < 0.001$
Hispanic, *N* = 7	34 (14.7, 10.6–53.1)	6.7 (9, 1.2–19.6)	$p = 0.009$
Asian, *N* = 2	29.2 (n/a, 21.7–36.6)	12.4 (n/a, 4.7–20.1)	$p = 0.009$
Other, *N* = 1	55.9	5.1	
Comorbidity			
Present, *N* = 57	28.1 (29.1, 5.5–178.9)	7.6 (6.6, 0.5–34)	$p < 0.001$
Absent, *N* = 49	31.5 (19.0, 6.6–72.7)	5.4 (4.9, 0.4–20.9)	$p < 0.001$
BMI percentile			
≥85, *N* = 87	29.8 (25.5, 5.5–178.9)	6.6 (5.9, 0.5–34)	$p < 0.001$
<85, *N* = 19	25.6 (17.2, 7.3–72.7)	6.1 (6.5, 0.4–25.1)	$p < 0.001$

TABLE 4 Residual OSA by category

Characteristic	Number (%)
Total	97 (91.5)
BMI percentile	
≥85, *N* = 87	81 (93.1)
<85, *N* = 19	16 (84.2)
Race	
African American, *N* = 75	70 (93.3)
White, *N* = 21	17 (80.9)
Comorbidities	
Present, *N* = 57	51 (89.5)
Absent, *N* = 49	46 (93.9)

These include African American race, high BMI, OSA severity, and presence of comorbidities such as asthma, Down syndrome, and craniofacial abnormalities.[23]

In our study, African American patients were more likely to experience residual OSA than white patients, although the majority of patients in both racial groups experienced residual OSA. Patients with medical comorbidities in this study were found to have a less dramatic reduction in oAHI; and although they experienced residual OSA at rates similar to those patients without medical comorbidities, this may be due to the fact that only a small number of patients experienced complete elimination of oAHI on postoperative PSG. More often, patients experienced objective surgical success, defined by an oAHI improvement by at least one diagnostic category of OSA. Cardiac abnormalities were the most common medical comorbidity found, followed by non-anatomic upper airway diseases, endocrine, and developmental disorders. Patients with developmental disorders had the least improvement in oAHI, followed by patients with cardiac disorders. This is consistent with literature that demonstrates higher rates of residual OSA in patients with neurologic disorders.[23]

Obesity is one of the greatest risk factors for developing OSA, with an incidence of up to 60% in obese children.[24] Additionally, obese children with OSA have been shown to have much more severe OSA, as demonstrated by higher mean oAHIs.[1,18,24] Although surgical success rates in this study were similar between obese and nonobese patients, patients in this study who were overweight or obese were less likely to reach a postoperative oAHI <5 or an oAHI reduction >85% after initial surgery.

With the potential for residual OSA or incomplete treatment, it is advantageous to determine which patients may benefit from concurrent surgical procedures, rather than an isolated AT. While characteristics described above may suggest a multifactorial cause of disease, DISE assists in identifying additional sites of obstruction that can be corrected. This study has demonstrated that performing DISE at the time of initial sleep surgery leads to significant improvements in postoperative PSG findings. A recent expert consensus statement has bolstered this finding, definitively reporting that patients undergoing DISE-directed surgery experience an improvement in OSA symptoms and quality of life, as well as improvements in AHI and oxygen saturation nadirs.[25] DISE also facilitates identification of collapse at locations that are difficult to view on physical exam alone. In pediatric patients, this can be because children often do not tolerate flexible laryngoscopy exams for a long enough period of time or simply because the location of collapse is subtle. Multi-level collapse is also exposed with DISE as the examiner is able to take note of each airway collapse location and continue looking for additional potential sites, sometimes altering surgical plans.[26] Furthermore, because the airway functions differently while asleep than awake, these dynamic changes

are not visible during the awake exam, regardless of how well the child may tolerate flexible laryngoscopy. In the adult literature, awake endoscopy has been shown to be a poor predictor of DISE findings, further illustrating the need for a method to examine the airway during sleep.[27] DISE accomplishes this by simulating physiologic sleep, providing a more accurate representation of the specific collapse, or collapses, the patient is experiencing at home. Once an obstruction is visualized, the surgeon can perform the necessary procedure to either reduce the disease burden or eliminate it altogether. Our study tracked the surgical success rate by determining how many patients had an oAHI reduction indicating an improvement by at least one diagnostic category of OSA (severe, moderate, mild, and no OSA). Notably, the majority of patients with severe or moderate preoperative OSA had postoperative oAHI values suitable for cessation of CPAP.

Upper airway collapse can occur in many different locations in the pediatric population, each with their own corresponding surgical treatment. The most common sites of obstruction contributing to pediatric OSA include adenoids, tonsils, retropalatal, base of tongue, and hypopharyngeal area.[28] Our patients followed this trend in that, aside from almost universal adenotonsillar hypertrophy, they experienced soft palate collapse, base of tongue obstruction, and obstruction at the epiglottis. Interestingly, the soft palate site of obstruction was recorded in 65.4% of patients in our first study, yet only 21.7% in this study. In addition, multilevel collapse occurred in only a minority of patients in this study (10.4%), compared with 23.1% in our original study. The larger sample size in the current study could help explain both of these discrepancies. Additionally, the different comorbidities between the studies, including developmental abnormalities, and differences in racial/ethnic background could have affected frequency of collapse sites observed.

Visualizing multilevel collapse with DISE at the time of the initial surgery allowed for intraoperative decisions to be made to address the additional obstructive sites. The surgeries used to address these obstructions include the following: pharyngoplasty, lingual tonsillectomy (base of tongue reduction), supraglottoplasty, and inferior turbinate reduction—each almost universally accompanied by AT. DISE allows for a more personalized surgical approach to each patient's specific anatomic collapse, which yields a more significant effect on symptom improvement. Identifying all obstructions prior to beginning surgery allows for operative planning to include all necessary procedures to prevent the child from having to undergo multiple operations. It is important to discuss reasons why a surgery would not be performed for an identified obstruction on DISE. For example, 32.1% of patients in this study were found to have lingual tonsillar hypertrophy yet only 20.1% underwent a base of tongue reduction. This is because some families expressed their decision to forego tongue base reductions at the time of initial surgery regardless of DISE findings. DISE has a large effect on surgical planning, with some studies reporting that its utilization changes operative decision making between 30 and 60.8% of the time, depending on the study.[29–31] Reducing the number of operations a child must undergo leads to a decreased risk of adverse effects from anesthesia and a decreased cost for the family and for the facility. Although DISE may prolong the single operation, our study found no complications or adverse effects related to DISE, extended anesthesia use, or concurrent procedures.

The data in this study shows that surgery guided by DISE was effective at reducing a child's OSA from severe, to mild and moderate levels. Although we found one study that was unable to find a significant objective improvement in patients' OSA after using DISE-directed surgery, their study design involved DISE only after primary AT had failed to resolve symptoms.[32] When comparing outcomes of our study with a systematic review of AT alone in surgically naïve patients by Friedman et al, our results produced similar measures of success. However, we believe that our results are more favorable compared with those of this systematic review when comorbidities, racial/ethnic backgrounds, starting AHI values, and average BMI are accounted for. For instance, even when considering the nine most complicated and comorbid populations in the systematic review by Friedman et al, our average AHI% reduction is still superior despite our population having a higher rate of obesity and comorbidities, higher mean BMIs, and higher preoperative AHIs.[33] As AT is the standard of care for pediatric OSA at the time of this writing, it is important to compare our findings with results of AT alone. A review by Galluzzi et al. which cited persistence of OSA in around one third of children with AT alone is difficult to compare with our study due in part to differing comorbidities. The only comorbidity included in the review by Galluzzi et al. was Down Syndrome in a relatively small number of cases. However, we believe that the success rates observed in our study with DISE-directed surgery warrant further investigation and comparison.[4]

Although 3–6 months may not allow for complete observation of recovery from surgery, the PSG findings clearly indicate an improvement in oAHI. Additional longitudinal PSG measurements would be helpful to more firmly establish long-term results and benefits. However, based on our previous study and the current study's results, DISE appears to offer a safe and effective solution to treating severe OSA and assists in preventing residual OSA. DISE has the capability to significantly reduce patients' OSA burden and decrease the occurrence of residual OSA in patients at highest risk for a poor response to standard first-line treatment. Should patients have characteristics that predispose them to developing residual OSA, such as severe OSA, obesity, or African American ethnicity, DISE may be offered adjunctively to determine what additional procedures are required to decrease the chances the residual OSA occurs.

Our study has several limitations. First, regarding the definition of residual OSA that was used, a majority of our patients were deemed to have residual OSA despite having experienced a significant oAHI reduction and improvement of symptoms. This finding was expected given that we used the most stringent definition. However, fewer patients would have experienced residual OSA had we used an oAHI cutoff of <5, or even <2, as seen in other studies described. Second, our study lacked long term PSG measurements. Our average postoperative PSG measurement occurred at around 4 months. More longitudinal measurements would provide evidence for the persistence of beneficial effects seen from DISE-guided surgery. Additionally, our study's demographics did not accurately reflect the population, with

African Americans comprising a majority of our sample. Herein lies a potential issue for generalizing our results to a broader population. Lastly, we did not record subjective clinical judgment of symptom improvement at follow up. This would have allowed a more holistic view of OSA improvement thanks to DISE-directed surgical decisions.

5 | CONCLUSION

This study supports the use of DISE to identify and treat obstructive lesions during initial surgery for severe sleep apnea in the pediatric population. DISE-directed intervention effectively aided in significantly reducing surgically naïve patients' mean oAHI. Patients with a lower BMI percentile were more likely to experience a postoperative oAHI <5. Patients with a higher BMI percentile were more likely to have residual OSA, although the difference was not significant. DISE allowed for additional sites of collapse to be identified and addressed concurrently with the patients' primary adenotonsillectomy. Future studies can investigate additional factors that contribute to long-term surgical success and persistence of results through more longitudinal PSG measurements. Additionally, a single, standardized definition of residual OSA should be clarified so that results can be more easily compared across studies and so that more effective methods for resolving patients' OSA can be discovered.

ORCID

Robert M. Frederick II https://orcid.org/0000-0001-6487-1567
Josiah Brandt https://orcid.org/0000-0001-8441-5630
Anthony Sheyn https://orcid.org/0000-0001-5924-3689

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