

Evolving Therapies to Treat Retroglossal and Base-of-Tongue Obstruction in Pediatric Obstructive Sleep Apnea

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Objective: To describe our experience treating retroglossal and base-of-tongue collapse in children and young adults with obstructive sleep apnea using combined genioglossus advancement (Repose THS; MedtronicENT, Jacksonville, Florida) and radiofrequency ablation of the tongue base.

Design: Retrospective institutional review board–approved analysis of 31 operations.

Setting: Tertiary pediatric medical center.

Patients: Thirty-one patients with a mean age of 11.5 years (age range, 3.1–23.0 years).

Interventions: Combined genioglossus advancement and radiofrequency ablation.

Main Outcome Measures: Preoperative and postoperative polysomnographic data were evaluated for each patient. Success of surgery was determined using the criteria of a postoperative apnea-hypopnea index of 5 or fewer events per hour, without evidence of hypoxemia

(oxygen saturation as measured by pulse oximetry), and without prolonged hypercarbia (end-tidal carbon dioxide).

Results: Thirty-one patients who underwent genioglossus advancement were analyzed. Nineteen (61%) had Down syndrome. The overall success rate was 61% (19 of 31) (58% [12 of 19] success among patients with Down syndrome and 66% [7 of 12] success among patients without Down syndrome). Overall, the mean apnea-hypopnea index improved from 14.1 to 6.4 events per hour ($P < .001$); the mean nadir oxygen saturation as measured by pulse oximetry during apnea improved from 87.4% to 90.9% ($P = .07$).

Conclusions: Pediatric obstructive sleep apnea refractory to adenotonsillectomy that is due to retroglossal and base-of-tongue collapse remains difficult to treat. However, most patients in this analysis benefited from combined genioglossus advancement and radiofrequency ablation.

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THE SPECTRUM OF SLEEP-related breathing disorder ranges in severity from primary snoring to obstructive sleep apnea (OSA). Approximately 1% to 3% of all children have OSA,¹ most of which is attributable to adenotonsillar hypertrophy. Patients who are severely affected may demonstrate craniofacial maldevelopment (adenoid facies), failure to thrive, delay in cognitive abilities, hypertension (pulmonary and systemic), or severe cardiovascular dysfunction, including cardiac failure from cor pulmonale.²⁻⁴

Fortunately, because most pediatric OSA is caused by adenotonsillar hypertrophy, adenotonsillectomy alone is an effective and durable treatment.⁵ However, multilevel airway narrowing beyond enlarged tonsils and adenoids may occur and

represents a source of treatment failure after adenotonsillectomy. Although these additional levels of narrowing (nasal, nasopharyngeal, retropalatal, retroglossal, and hypopharyngeal)⁶ may be found in otherwise healthy children, certain populations are predisposed to multilevel airway collapse. Included in this group are obese children, children with nasal obstruction of any etiology, children with neurologic impairments or familial factors, children with malacia or laryngotracheal or bronchial stenosis, and children with craniofacial anomalies, such as Pierre Robin sequence and Down syndrome.⁷

The treatment of pediatric OSA beyond adenotonsillectomy is more complex. Managing OSA of anatomically diverse origins includes diagnosing which levels are responsible for the airway collapse and defining the severity of the col-

lapse. A polysomnogram (PSG) is used to assess the severity of the collapse.⁷ Although most clinicians do not routinely obtain a PSG in children with OSA and with classic signs and symptoms of adenotonsillar hypertrophy, multilevel collapse (especially after adenotonsillectomy) needs to be quantified using a PSG before proposing more invasive surgical treatments or continuous positive airway pressure (CPAP) management.

Diagnosing which levels are responsible for airway collapse can be difficult. Included in this workup are a detailed history and physical examination, including flexible nasopharyngoscopy to the level of the larynx.⁸ Recently, cine magnetic resonance (MR) imaging has provided a useful radiographic adjunct to the physical examination, because it allows the clinician to screen for and to observe airway collapse in 3 planes (axial, coronal, and sagittal).^{9,10} The Bernoulli principle indicates that collapse at one level may mask or enhance collapse at another location in the airway. Therefore, the exact nature of dynamic airway collapse may not be appreciated by a detailed history and physical examination, cine MR imaging, and flexible endoscopy performed in the office and in the operating room.

Once OSA following adenotonsillectomy has been confirmed by a PSG and the levels of collapse have been elucidated, the treatment options are surgical and nonsurgical. Much pediatric OSA could be ameliorated with CPAP, which avoids surgery. The necessary amount of positive pressure is determined during a special CPAP titration PSG, and CPAP is typically delivered using a face mask, a nasal mask, or nasal pillows. Realistically, the success of CPAP treatments is diminished by poor patient compliance. One-third of adult patients opt out of CPAP therapy when required for persistent OSA after adenotonsillectomy.¹¹ Reasons for noncompliance include the obtrusiveness of the machine, a sense of claustrophobia, or physical discomfort.

For these reasons, adult and pediatric patients with OSA frequently prefer a surgical option if one is available. The objective of the present study is to describe our experience performing combined genioglossus advancement (GGA) (Repose THS; Medtronic ENT, Jacksonville, Florida) and radiofrequency ablation (RFA) of the tongue base to treat OSA in children that is refractory to adenotonsillectomy and is attributable to retroglossal and base-of-tongue collapse.

METHODS

Among 31 patients with OSA persisting after adenotonsillectomy who were thought to have obstruction at the base of the tongue, a retrospective analysis was conducted to define postoperative success after combined GGA and RFA (n=30) or GGA alone (n=1). Patients undergoing lingual tonsillectomy, revision adenoidectomy, or uvulopalatoplasty with simultaneous GGA were included. Patients undergoing RFA or lingual tonsillectomy without sequential or simultaneous GGA were excluded from this study. Likewise, patients undergoing GGA who lacked a preoperative or postoperative full-night PSG, patients whose PSG was conducted with supplemental oxygen, and patients whose PSG was for CPAP titration were excluded. This study was approved by the institutional review board.

POLYSOMNOGRAPHY

Criteria for success were based on a postoperative PSG. As suggested in a recent update by Halbower et al,¹² a successful outcome was defined as (1) an apnea-hypopnea index (AHI) of 5 or fewer events per hour, (2) oxygen saturation as measured by pulse oximetry (SpO₂) of at least 90%, and (3) total sleep time spent with end-tidal carbon dioxide (ETCO₂) exceeding 50 mm Hg of less than 10%. The following definitions were used to score PSGs: *Obstructive apnea* occurs if there is an absence of airflow for 2 or more respiratory cycles but the abdominal and thoracic movements continued. *Mixed apnea* occurs if airflow and abdominal and thoracic movements stop but are followed by abdominal and thoracic movements without airflow for at least 2 respiratory cycles. *Central apnea* is defined as the cessation of airflow and abdominal and thoracic movements. Central apnea is clinically significant if it is associated with oxygen desaturation exceeding 4%, if it is longer than 20 seconds, or if it is associated with bradycardia. *Hypopnea* is defined as a 50% reduction in airflow. Hypopneas are also associated with a 4% decrease in oxygen saturation or with an arousal that follows hypopnea. The *respiratory disturbance index* is the number of central, obstructive, or mixed apneas plus hypopneas per hour of sleep. The *AHI* is the number of obstructive apneas and hypopneas and the number of mixed apneas and hypopneas that occur per hour of sleep. In children, a normal AHI should be less than 1.0. Obstructive sleep apnea is defined using the AHI, with 1 to 5 indicating mild OSA, greater than 5 to 15 indicating moderate OSA, and greater than 15 indicating severe OSA.

SURGICAL PROCEDURES

Genioglossus advancement was performed according to the manufacturer's directions and included placing a titanium screw with a preattached heavy polyethylene suture into the genial tubercle through a submental incision. The heavy suture was then passed through the tongue base in a triangular fashion and was tied to the opposite suture near the genial tubercle. The tightness of the knot was titrated based on simultaneous palpation of the tongue base.

Radiofrequency ablation was performed as described by Powell et al.¹³ In brief, 1.5 mL of 1:200 000 bupivacaine hydrochloride was injected into the tongue base before RFA into sites where the probe would be inserted. A 2-pronged probe was used at a setting of 600 J. The probe was initially inserted at the vertex of the circumvallate papillae. Subsequent lesions were created 1 cm anterior to the first treatment site and, as indicated, 1 cm anterior to the second treatment site.

A plasma wand (XP Coblation; ArthroCare ENT, Austin, Texas) was used to perform lingual tonsillectomy on patients with lingual tonsillar hypertrophy who were simultaneously undergoing combined GGA and RFA.¹⁴ The anterior midline of the tongue was controlled with a heavy silk suture, while an appropriately sized Lindholm laryngoscope (Karl Storz, Tuttlingen, Germany) was used to expose the tongue base. The plasma wand was used to remove lingual tonsillar tissue using settings 9 (ablate) and 5 (coagulate).

RESULTS

Thirty-one patients underwent GGA for OSA refractory to adenotonsillectomy. Only 1 patient underwent GGA as an isolated procedure, while the remaining 30 patients underwent combined GGA and RFA of the tongue base. Five patients underwent simultaneous lingual ton-

sillectomy. In 5 additional patients, other levels of obstruction were treated simultaneously with GGA and RFA: 3 underwent revision adenoidectomy for nasopharyngeal obstruction, and 2 underwent uvulopalatoplasty for retropalatal collapse. The mean age of these patients was 11.5 years (age range, 3.1-23.0 years), with 22 male patients and 9 female patients included in the study. Because of the retrospective nature of the study, body mass index and Mallampatti scores were unavailable. To determine the anatomical level of persistent airway obstruction, 28 of 31 patients had undergone cine MR imaging using the method described by Shott and Donnelly,¹⁰ which attributed the major site of obstruction to the tongue base. Sixty-one percent (19 of 31) of patients undergoing combined GGA and RFA had Down syndrome.

Surgical success was based on PSG results (**Figure**). The PSGs were obtained, on average, 5.6 months (range, 2-24 months) after surgery. The mean (SD) preoperative and postoperative percentages of sleep time spent in rapid eye movement were 16.2% (7.0%) and 15.6% (6.5%), respectively. The mean (SD) arousal index decreased from 15.0 (8.6) before surgery to 11.6 (5.8) after surgery ($P=.07$). The mean (SD) nadir SpO₂ during hypopnea rose from 86.1% (5.4%) before surgery to 89.1% (7.2%) after surgery ($P=.003$). Similarly, the mean (SD) nadir SpO₂ during apnea improved from 87.4% (8.6%) before surgery to 90.9% (5.9%) after surgery ($P=.07$). The mean (SD) AHI improved from 14.1 (10.1) events per hour before surgery to 6.4 (6.5) events per hour after surgery ($P<.001$) (Figure). The postoperative course was similar to that seen after adenotonsillectomy, with no patient having persistent dysphagia beyond 2 weeks after surgery. Because of 2 episodes of seroma in the immediate postoperative period, the technique was altered to include intraoperative placement of a drain in the submental incision, which was removed before discharge home.

A successful outcome consisted of maintaining oxygen saturation at or above 90% for the duration of the study, reducing the AHI to 5 or fewer events per hour, and keeping the total sleep time spent with ETCO₂ exceeding 50 mm Hg to 10% or less. According to these criteria, 16 patients had a successful outcome. Three additional patients were judged to have an excellent outcome based on parental assessment; these patients met the criteria for a successful outcome based on ETCO₂ and oxygen saturation, but their postoperative AHIs were between 5 and 6. These patients were also deemed to have successful outcomes, contributing to an overall success rate of 61% (19 of 31). Surgical success was achieved in 58% of children with Down syndrome (12 of 19) and in 66% of patients without Down syndrome (7 of 12). There was no statistically significant difference in success rates between patients with vs without Down syndrome who underwent combined GGA and RFA ($P=.45$, Fisher exact test).

COMMENT

Most pediatric OSA is attributable to adenotonsillar hypertrophy, and adenotonsillectomy alone is an effective

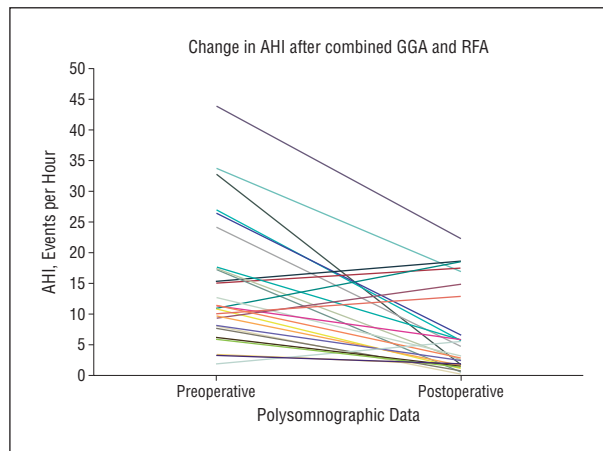


Figure. Preoperative and postoperative values for the apnea-hypopnea index (AHI) of 31 patients undergoing combined genioglossus advancement (GGA) (Repose THS; MedtronicENT, Jacksonville, Florida) and radiofrequency ablation (RFA) of the tongue base.

and durable treatment. In certain populations, multi-level airway obstruction is more common, and these patients demonstrate OSA that is refractory to adenotonsillectomy. The initial steps in treating children with OSA that is refractory to adenotonsillectomy include quantifying the severity of OSA with a full-night PSG and determining the levels of collapse through a detailed history and physical examination, flexible endoscopy in the office and in the operating room, and cine MR imaging.^{9,10} Patients with retroglossal airway obstruction after adenotonsillectomy may be candidates for CPAP, operative management, or both. Herein, we reviewed our experience with 31 consecutive patients undergoing combined GGA and RFA in whom retroglossal collapse was thought to be the primary site of obstruction. Using strict postoperative polysomnographic criteria, this surgical approach was successful in most patients.

Several operations have been developed to improve retroglossal collapse. Radiofrequency ablation provides a minimally invasive means to deliver limited thermal damage to the tongue base, creating lesions that diminish the bulk and flaccidity of the tongue base through fibrosis. The GGA technique attempts to stabilize the tongue base to prevent its retrodisplacement during supine sleep. Differing from musculoskeletal advancement procedures, GGA relies on a heavy suture that is triangulated through the tongue base submucosally to apply tension directly at the site of obstruction. Over time, a fibrotic bridge forms around the implanted suture and provides additional strength.

Other surgical procedures that address base-of-tongue obstruction described in adult populations include hyoid myotomy and suspension, as well as midline mandibular osteotomy with GGA. The hyoid myotomy and suspension procedure advances the hyoid complex to improve the retroglossal airspace by placing traction on the hyoid directly. The midline mandibular osteotomy procedure pulls the genioglossus muscle forward from its insertion onto the genial tubercle. Both of these procedures have shown variable success in adults¹⁵ and are of limited use in children because of the chang-

ing position of the hyoid as a child grows. Midline mandibular osteotomy also may affect secondary tooth buds in children.

Patients who fail these phase 1 surgical interventions or in whom severe retrognathia is present may be candidates for maxillomandibular advancement surgery. In this technique, the retropalatal and retroglossal airway is increased by performing a Le Fort I maxillary osteotomy and sagittal split mandibular osteotomies with advancement.¹⁶ Although tracheotomy bypasses the retroglossal airway, most practitioners prefer to reserve tracheotomy for children with severe affliction.

The pediatric OSA literature reports limited experience in applying GGA. In the adult OSA literature, a review of 14 patients aged 35 to 74 years undergoing GGA demonstrated a decrease in the mean respiratory disturbance index from 35 to 17, as well as improved scores on a nonvalidated questionnaire as rated by their partners at 2 months.¹⁷ In another prospective study, Terris et al¹⁸ evaluated 19 adults (mean age, 44.9 years) with OSA who underwent GGA. Twelve of 19 had postoperative PSGs, and the mean AHI decreased from 32.4 before surgery to 14.4 after surgery. The AHI and Epworth sleepiness scores also improved. However, a recent meta-analysis of GGA that includes these results found an overall success rate of 20% to 57%.¹⁷ Because of widely differing outcomes criteria, it is difficult to compare our results in a pediatric population with these reported in adult studies.

To date, our results represent the first significant review of GGA applied to a pediatric OSA population. Using more stringent PSG criteria than those applied in the adult studies, we demonstrated an overall success rate of 61% (19 of 31) among patients undergoing combined GGA and RFA. Based on changes in the mean AHI, the mean nadir SpO₂ during apnea, and the mean percentage sleep time with ETCO₂ exceeding 50 mm Hg (Figure), statistically significant improvements were noted after surgery across all variables. Overall success was unaffected by simultaneous lingual tonsillectomy ($P = .66$, Fisher exact test).

Our data support the role of combined GGA and RFA in a pediatric population with OSA that is refractory to adenotonsillectomy. In a study by Schaaf et al,¹⁹ sagittal cine MR imaging was used to evaluate tongue size and bony confines of the oropharynx and hypopharynx in 28 children undergoing GGA. These children represent a subgroup of the population presented herein. The relative size of the tongue (ratio of the tongue to bony confines) was larger in operative successes than in failures (mean, 0.51 vs 0.46; $P = .02$). Smaller adenoids were also associated with operative successes (mean, 9.1 vs 12.4 mm; $P = .049$). No other biomarkers, including absolute tongue and airway size or dynamic airway motion, were significant. If combined GGA and RFA is most effective in the setting of relative macroglossia because of combined radiofrequency debulking and suture suspension of the tongue base, then operations that directly debulk the tongue base such as midline posterior glossectomy¹⁴ may provide more benefit.

Analysis of treatment success in pediatric OSA can take many forms depending on the outcomes measure used.

The present study defines success and failure based on PSG variables, including AHI, SpO₂, and ETCO₂. The adult literature considers an AHI of 20 events per hour to be a reasonable threshold to treat; however, the pediatric literature uses more restrictive AHI values. Although an AHI exceeding 1 event per hour is considered abnormal by many pediatric sleep medicine practitioners,^{12,20} it would be unreasonable to recommend a tongue base operation to such a patient without a nadir SpO₂ below 90% or without significant sleep time spent in hypercapnia. The present review considers 5 events per hour to signify patients who might benefit from surgery. However, 3 patients herein with AHIs between 5 and 6 after undergoing GGA were deemed surgical successes because they had no concomitant hypercapnia or desaturation and their sleep architecture had subjectively and objectively improved. The preoperative AHIs for these 3 patients were 11.3, 26.5, and 26.9 events per hour. Likewise, patients who were CPAP intolerant before surgery who became CPAP tolerant after surgery (regardless of PSG results) represent some degree of improvement, although we did not use CPAP tolerance as a criterion for success in the present study.

As with any retrospective analysis, a major limitation of our study was the inability to control the data. In our series of 31 consecutive patients undergoing combined GGA and RFA, few patients underwent simultaneous lingual tonsillectomy. However, the reported success rates with GGA are not altered statistically by lingual tonsillectomy, nor were the success rates altered statistically by the addition of revision adenoidectomy or uvulopalatoplasty in 5 additional patients. Likewise, although children with Down syndrome represented a physiologically unique group in our study, no statistically significant difference was seen in overall success rates between patients with vs without Down syndrome undergoing combined GGA and RFA. Finally, PSGs were performed no less than 2 months (range, 2-24 months) after surgery. However, it is unclear what the optimal period should be for a postoperative PSG. In 2 patients, we observed improvement over time (based on a second postoperative PSG) without any additional operations. In both of these patients, parental reports of improvement led to a second postoperative PSG. For children who failed to improve sufficiently following combined GGA and RFA, most are being treated with CPAP or with bilevel positive airway pressure.

In conclusion, patients with retroglossal and base-of-tongue obstruction after adenotonsillectomy may be candidates for CPAP or for operative management that includes combined GGA and RFA. Our results indicate the combination of GGA and RFA is successful in alleviating pediatric OSA in most appropriately selected patients. However, because we theorize that it is a combination of glossoptosis and macroglossia that contributes to retroglossal collapse, procedures that more aggressively reduce the bulk of the tongue base may be indicated. Accordingly, we are enrolling patients into a prospective study of refractory pediatric OSA that sequentially applies midline posterior glossectomy with plasma wand ablation, followed by combined GGA and RFA (if necessary).

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