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The Role of Lingual Tonsillectomy in Treating Pediatric Chronic Upper Airway-Related Symptoms

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ABSTRACT

Introduction: Lingual tonsil hypertrophy (LTH) is a possible cause of childhood obstructive sleep apnea after adenotonsillectomy, especially in patients with trisomy 21, but its role in chronic cough and exercise induced shortness of breath in non-syndromic patients is less well-established. As such, lingual tonsillectomy is rarely performed. This study is a preliminary exploration of the role of lingual tonsils and subsequent lingual tonsillectomy in pediatric patients with chronic respiratory-related symptoms refractory to more traditional therapies.

Methods: Twelve children were referred by a pediatric pulmonologist with chronic upper airway symptoms unresponsive to traditional treatments, and for whom the only abnormal finding on flexible bronchoscopy was LTH. Endoscopic lingual tonsillectomy was subsequently performed, and the post-operative response was assessed.

Results: All patients saw preoperative symptom resolution one month after surgery. One patient experienced symptom recurrence and LTH again after one year but has seen symptom resolution 14 months after surgical revision. One patient subsequently underwent palatine tonsillectomy due to symptom persistence two years later. There were no intraoperative complications and only one patient experienced mild and self-limited pharyngeal bleeding eight days postoperatively.

Conclusion: Lingual tonsillectomy proved to significantly resolve chronic respiratory related symptoms when no other treatment had in this case series, implicating LTH in difficult-to-treat symptoms, including chronic cough, throat-clearing and shortness of breath. Though sample size is a limitation, this study suggests that the role of the lingual tonsils should not be overlooked in patients with these types of symptoms.

Keywords: Pediatric; Lingual tonsil; Hypertrophy; Upper-Airway Obstruction; Tonsillectomy

INTRODUCTION

Tonsils are a collection of B-cell lymphoid tissue that plays an important role in immune defenses due to their positioning prior to the digestive and respiratory tracts.1 The oropharyngeal tonsils compose Waldeyer's ring and

consist of palatine, pharyngeal (adenoid), lingual, and tubal.2 The tonsils grow to completion between the 6th to 8th year of life, though are the most immunologically active between the 4th and 12th years, after which they begin to atrophy [1].

The lingual tonsils are located at the base of the tongue anterior to the epiglottis, posterior to the circumvallate papillae, and medial to both tonsillar pillars [2]. They are arterially supplied by the lingual branch of the external carotid artery, the lingual veins of the internal jugular vein carry blood away, and they are lymphatically supplied by the superior deep cervical lymph nodes [1].

Though it remains to be rarely performed, lingual tonsillectomy has gained popularity in recent years due to either surgical innovations, such as video laryngoscope and microdebrieder resection, or increased knowledge of the lingual tonsils' contribution to diseases [3]. Lingual tonsillar hypertrophy (LTH) can occur in the presence of acute or chronic inflammation, such as alcohol, tobacco, allergic rhinitis, elevated BMI and laryngopharyngeal reflux, and lead to airway obstruction [4]. Additionally, prior removal of other components of Waldeyer's ring, both with and without adequate indication, has been theorized to contribute to compensatory lingual hyperplasia [5,6]. (The cause of lingual tonsillitis has been shown to be similar to the etiologies of pharyngitis and adenotonsilits, such as group strep A and staphylococcus aureus. In a study of 602 adult patients who underwent lingual tonsillectomy, indications were 58.7% for OSA, 18.8% LTH and infection, and 15.9% for neoplasia [7]. Though lingual tonsillectomy has a therapeutic effect for all age groups, one of its most prominent roles is in the treatment of refractory obstructive sleep apnea (OSA) in pediatric populations [8]. First-line treatment of OSA remains nonsurgical in the form of weight loss and continuous positive airway pressure in adults, as the latter is generally poorly tolerated in children.

Therefore, surgical intervention via adenotonsillectomy is the first-line treatment considered in children. Studies have shown that 20 to 40% of children continue to have post-operative airway obstruction contributing to OSA, elucidating the role that other factors, including LTH, most likely plays in OSA's pathological disease process [9]. Children with a prior diagnosis of Down Syndrome have a higher likelihood of developing refractory OSA, and LTH should be considered as an underlying etiology of airway obstruction. Additional reasons for OSA persistence in this population are muscular hypotonia, hypoplasia and hypopharyngeal collapse [10]. In general, lingual tonsillectomy across all age groups lacks significant morbidities, adverse events are relatively rare, and post-operative readmission rates are similar to those of other types of tonsillectomies [3,11]. However, evidence suggests that children with comorbidities such as Down Syndrome, neurological syndromes, craniofacial disorders, and obesity (>85 percentile BMI) are more likely to have poor surgical outcomes and higher postoperative AHI after lingual tonsillectomy [9,10]. This data is limited and additional studies are required to further investigate this.

Due to its success rate in treating refractory OSA, there is thought to be an indication for lingual tonsillectomy in children with other forms of airway obstruction. Twelve patients with a history of chronic cough and throat clearing while playing sports underwent pulmonary flexible bronchoscopy under anesthesia and were found to have no underlying etiologies, apart from LTH. In our study, we evaluate the role of lingual tonsillectomy in children with chronic cough and throat-clearing, particularly during activity, refractory to standard treatment methods.

MATERIALS AND METHODS

A retrospective chart review was performed for all patients who underwent lingual tonsillectomy for lingual tonsil hypertrophy (LTH) from 2018 to 2022 in a tertiary care pediatric otolaryngology practice. A total of 12 patients were identified. The study was approved by the NOVA Southeastern College of Allopathic Medicine (NSU IRB Protocol Number 2022-302). Mean follow-up time for all patients included in the present study was Follow-up is currently ongoing.

Patient Population: Patient data, including age, sex, past medical history, upper airway-related symptoms (indications for surgery), tonsillar size, and pre-surgical treatments are shown in Table 1. All patients enrolled were 18 years or younger and under the care of a pediatric pulmonologist for upper airway-related symptoms, including cough, throat-clearing, exercise-induced shortness of breath, and snoring for at least 6 months. Some patients also had a sensation of solid foods becoming stuck in the back of the throat when swallowing. The differential diagnosis for patients included laryngopharyngeal reflux, allergic rhinitis, vocal cord dysfunction, and reactive airway disease, and patients were treated medically according to their symptoms with no resolution. All patients were found to have lingual tonsil hypertrophy on flexible bronchoscopy and referred to otolaryngology for lingual tonsil removal.

Table 1: Patient Demographics and Medical History.

Patient	Sex at birth	Age at Surgery	Past Medical History	Past Surgical History
1	Male	15	RA, GERD, GP	T&A, NA
2	Male	10	PANDAS, ADHD, GERD, MFTHR gene	T&A, EGDx2
3	Female	16	VWD T1, GERD, BP	T&A, NS
4	Female	17	RSTIx2 years	-
5	Male	8	Narcolepsy, ROM	T&A, BMT
6	Female	15	Allergies	-
7	Male	15	CD	Colonoscopy
8	Male	8	-	-
9	Male	18	GERD	-
10	Male	17	RAD	-
11	Male	5	Concussion, AR	A
12	Male	16	CE, RAD	EGD

RA, rheumatoid arthritis; GERD, gastroesophageal reflux disease; GP, gastroparesis; PANDAS, Pediatric Autoimmune Neuropsychiatric Disorders Associated with Streptococcal Infections; ADHD, attention-defecit hyperactivity disorder; MFTHR, methylenetetrahydrofolate reductase; VWD T1, Von Willebrand Disease Type 1; BP, bipolar disease; RSTI, recurrent strep throat infection; CD, celiac disease; RAD, reactive airway disease; AR, allergic rhinitis; CE, chronic esophagitis; T&A, tonsillectomy and adenoidectomy; NA, needle aspiration of knee; EGD, Esophagogastroduodenoscopy; NS, nasal surgery; ROM, recurrent otitis media; BMT, bilateral myringotomy with tubes; A, adenoidectomy

Pre and Postoperative Assessment: Preoperative assessment of patients included patient and parent/guardian self-report of symptoms before and after trial of mediation (Table II). Further patient assessments sometimes also included sleep study, spirometry, and upper endoscopy (Table III). A CT scan was performed on only one patient. In 6 patients, palatine tonsil size was noted and reported on physical exam and adenoid size was measured via in-office endoscopy, as all others had undergone prior adenotonsillectomy. Flexible bronchoscopy was performed

by the pediatric pulmonologist for all patients. Preoperative bronchoscopy was performed within 6 months of surgery.

Postoperative assessment primarily included patient and parent/guardian self-report of symptoms. Flexible endoscopy or bronchoscopy and diagnostic sleep endoscopy were performed in select patients who experienced persistence of symptoms. Patients were followed postoperatively. The duration of follow-up was recorded. The presence of symptoms was rated on a scale of 0 to 1, or no symptoms to the presence of symptoms, respectively.

Indications for Surgery

The following conditions were considered for surgery:

- Upper airway-related symptoms lasting more than 6 months
- Failure of conservative treatment in the form of medication for more than 6 months
- History of negative upper airway imaging and specialized testing
- Requirement for lingual tonsil hypertrophy as the only positive finding on flexible bronchoscopy

Surgical Method: Lingual Tonsillectomy: After informed consent was signed, the patient underwent general anesthesia, in supine position, with nasotracheal intubation. A dose of IV dexamethasone was given (4 mg). The tip of the tongue was grasped with an Allis clamp, and the tongue was retracted outward from the oral cavity while a McIvor mouth retractor was placed. After the mouth retractor was opened, the Allis clamp was removed, and the mouth retractor was placed into suspension. A red rubber catheter was placed trans-nasally (on the opposite side of the nasotracheal tube) and retrieved trans-orally and used to retract the soft palate. The lingual tonsils were then visualized using a 4mm Hopkins rod rigid 30-degree telescope (Karl Storz, Tuttlingen, Germany), attached to a light source, camera, and TV monitor. The lingual tonsils were then removed using radiofrequency ablation (Procise Max, Smith+Nephew, Andover, Massachusetts, USA). The lingual tonsils were removed on an ablation setting of 8. The anterior boundary was just posterior to the circumvallate papillae; the posterior boundary was the lingual surface of the vallecula, the lateral boundaries were at the junction of the inferior pole of the palatine tonsil on each side, and the depth was to the smooth investing fascia of the base of the tongue. Care was taken to not violate the tongue base fascia (and thus not to expose any tonsil musculature). Also, when working in the vallecula (midline), care was taken to retract the lingual tonsil tissue anteriorly with the device before activating the ablation, to avoid mucosal injury to the mucosal of the lingual surface of the epiglottis. After surgery, patients were observed for 2 hours, and if doing well from a respiratory and oral feeding status standpoint, were sent home, with instructions to take ibuprofen and/or acetaminophen as needed for pain. If ≥12 years old, a prescription for acetaminophen/hydrocodone was given, to be used only as needed.

STATISTICAL ANALYSIS

Data were entered into an Excel 2018 (Microsoft, Redmond, WA) spreadsheet and analyzed using SPSS statistical software (IBM, Armonk, NY). To determine if lingual tonsillectomy resolved patient symptoms postoperatively, a 95% confidence interval was performed. This was performed three times for resolution of symptoms 1 month, 1 year, and 2 years postoperatively. All patients were included in the first calculation. 1 patient was excluded in the second, and 3 patients were excluded in the third based on follow-up times.

RESULTS

Twelve patients were enrolled and underwent lingual tonsillectomy for treatment of upper airway-related symptoms. 75% of the patients were male and 25% were female with an average age of 13.3 (range: 5 to 19) years old at the time of surgery (Table 1). Upper airway-related symptoms primarily included shortness of breath with some relation to exercise (50%), snoring (33%), and recurrent choking on food (33%) (Table 2). Average duration of patient symptoms was 25.25 months with a lower limit of 6 months and an upper limit of 120 months. Incidentally, 33% of patients were also found to snore at night, with only patient 5's being severe. 33% of patients had previous tonsillectomy with adenoidectomy and one patient had previous adenoidectomy. The remaining patients had +1 tonsil size and small adenoids on in-office endoscopy (Table 3). Table II presents the diagnoses given to the patients prior to bronchoscopy and the specific treatments provided.

Table 2: History of Patient Symptoms and Treatment.

Patient	Upper-airway symptoms	Duration of Symptoms	Prior Diagnosis	Prior Treatment
1	EISOB, D with food	12 months	SOB, D	PPI
2	SOB, CC, D with food	8 months	SOB, D, CC	AHx2, Steroid (IN), Antacidx2
3	SOB	6 months	SOB	AH, Antacid
4	EISOB, D with food	120 months	D	AH, Antacid
5	Snoring, persistent OSA	12 months	OSA	-
6	Snoring, EITT	15 months	SOB, GS	H2B, Steroid (IN)
7	EISOB	12 months	EISOB	AH, Antacid, Steroid (IN), AI
8	Snoring, CC, Choking at night	10 months	CC, SDB	AHx2, Antacid, Steroid (IN), AI, Steroid (PO)x3, ABx3
9	Choking at night	12 months	OSDB	PPI, AH, Antacid, Steroid (IN), AI, BDx3
10	Snoring, EISOB Fatigue	12 months	EISOB	AH, Antacid
11	TC	12 months	CTC	H2B, Steroid (IN), Steroid (PO), BD,AI
12	EISOB, GS	72 months	EISOB, GS	AH, PPI, H2B, Steroid (IN)

EI)SOB , (exercise-induced) shortness of breath; D, dysphagia; CC, chronic cough; GS, globus sensation; VCD, vocal cord dysfunction; (O)SDB, (obstructive) sleep disordered breathing; EITT, exercise-induced throat tightness; TC, throat clearing; CTC, chronic throat clearing; PPI, proton-pump inhibitor; AH, antihistamine; H2B, H2-Blocker; AB, antibiotics, AI, antiinflammatory inhaler; IN, intra-nasal route of administration; PO, oral route of administration; BD, bronchodilator.

Table 3: Patient Examination Findings.

Exam	Patient	Result
Physical exam for tonsils*		
	4	size +1
	6	size +1
	7	size +1
	8	size +1
	9	size +1
	10	size +1
	11	size +2
	12	size 1+
In-office endoscopy for adenoids**		
	4	small
	6	small
	7	small

	8	small (15%)
	9	small
	10	small (20%)
	12	small (10%)
Spirometry		
	2	negative findings
	6	negative findings
	10	upper airway obstruction
Upper endoscopy		
	4	negative findings
CT Neck		
	9	negative findings
Flexible Bronchoscopy		
	10	Lingual tonsil hypertrophy
Paranasal Sinus X-Ray		
	11	Left maxillary chronic sinusitis

*Patients 1, 2, 3, 5 had previous tonsillectomy

**Patients 1, 2, 3, 5, 11 had previous adenoidectomy

Patient 2 and 5 had previous diagnoses of ADHD and narcolepsy controlled on medication, respectively. Patient 4 was also treated with 3 courses of antibiotics for recurrent strep-throat infections 1 to 2 times per year over 2 years. Other comorbidities include Rheumatoid Arthritis with chronic rheumatoid lung nodules, PANDAS, MFTHR gene, Type 1 Von Willebrand Disease, Bipolar Disorder, Obstructive Sleep Apnea, Celiac Disease, gastroesophageal reflux disease, recurrent otitis media, and reactive airway disease. Other than tonsillectomy and adenoidectomy, other surgical history included needle aspiration of the knee, esophagogastroduodenoscopy, multiple endoscopy nasal septal cautery surgeries, endoscopic excision of aright posterior nasal cavity pyogenic granuloma, bilateral myringotomy with tubes, and colonoscopy (Table 2).

Select patients also underwent further testing and imaging. Patients 2, 6 and 10 underwent spirometry with the pulmonologist, though only patient 10 had upper airway obstruction. Patient 4 had an upper endoscopy, patient 9 had a CT scan and patient 10 had a sleep study, all of which had negative findings. Patient 11 underwent a paranasal sinus x-ray which was positive for left maxillary chronic sinusitis. Despite some cobblestoning of the hypopharynx in patient 8, the only finding on flexible bronchoscopy, and a requirement for surgical lingual tonsil removal, was lingual tonsil hypertrophy (Table 3).

In all patients, there were no intraoperative complications and no patients required readmission after surgery. There were two postoperative complications in patient 10 and 11. On day three, patient 11 experienced a brief syncopal episode due to dehydration. On day eight, patient 10 experienced brief, mild and self-limited pharyngeal bleeding, and dark stools. At all the time points, patients preoperatively reported having symptoms. Of the 12 patients evaluated after 1 month, no patient reported symptoms. Of the 9 patients evaluated after 1 year, 1 patient reported recurrence of symptoms. Patient 9 experienced symptom recurrence 9 months postoperatively due to palatine tonsillar obstruction seen on follow-up bronchoscopy and diagnostic sleep endoscopy, and palatine tonsillectomy and adenoidectomy was performed 6 months later. Of the 7 patients evaluated after 2 years, 1 patient reported symptom recurrence due to lingual tonsillar regrowth and recurrence of hypertrophy seen on in-office flexible endoscopy with the pediatric otolaryngologist Patient 1 has been symptom-free following revision lingual tonsillectomy performed 14 months ago. At all the time points, lingual tonsillectomy for the entire cohort as a

group was shown to significantly improve patient symptoms. The 95% confidence interval estimating the percentage success at each time point and the margin of error is displayed in Table 4. The only limitation of this study is in patient 11, who underwent concomitant endoscopic left maxillary antrostomy and bilateral inferior nasal turbinate submucosal resection with lingual tonsillectomy.

Table 4: Presence of Symptoms Pre and Post Lingual Tonsillectomy.

Number of Months Postoperative	n	Number of Patients with Symptoms	95% CI
1	12		UB: 100%
		0	LB: 78%
12	9		UB: 100%
		1	LB: 54%
24	7		UB: 99%
		1	LB: 46%

DISCUSSION

The etiology of LTH is not clear, but has previously been linked to laryngopharyngeal reflux, high BMI, allergic rhinitis, and younger age.5,6 Compensatory enlargement of lingual tonsils after childhood adenotonsillectomy has also been commonly reported in the literature [5,12]. We also know that LTH has increased prevalence among pediatric patients with down syndrome due to studies such as the case-control lingual tonsil size analysis performed by Sedaghat et al. [13].

LTH has been cited numerous times in the literature as a possible cause of OSA that is refractory to adenotonsillectomy, as lingual tonsillectomy has provided OSA improvement in pediatric patients [3,9,10]. Lingual tonsillectomy, albeit an uncommonly performed procedure, has been implemented in children with persistent OSA despite adenotonsillectomy. The procedure has been reported as safe, with readmission and bleeding rates that are comparable to previously reported rates in conventional tonsillectomy [11]. LTH has also been presented in the literature as a potential problem in anesthesia, about tracheal intubation and face mask ventilation [14,15].

However, in line with the understanding that OSA is an upper airway concern, and with the previous evidence that lingual tonsillectomy improves OSA, LTH may be linked to a wider array of chronic upper respiratory issues. When enlarged, the lingual tonsils can displace the epiglottis posteriorly against the pharyngeal wall, allowing for the potential obstruction of air, food, or liquid through the nasopharynx and oropharynx [16,17].

Therefore, beyond obstructive sleep-disordered breathing, LTH may be implicated in exercise-induced and non-exercise-induced airway symptoms such as shortness of breath, throat tightness, throat clearing, dysphagia, cough, globus sensation, and snoring. To our knowledge, only a few case reports have considered the role of lingual tonsils in airway issues aside from OSA. In a case report by Lewis et al., they utilized lingual tonsillectomy on a pediatric patient with LTH and a chronic paroxysmal cough which was refractory to multiple attempts at medical treatment. Two weeks after the surgery he was no longer suffering from the cough and had no recurrence even at the 1-year follow-up.18 In another case report by Mowry et al., a lingual tonsillectomy performed for LTH resolved a 6-month history of worsening dysphagia in a pediatric patient, who had been evaluated and treated for many other possible causes of dysphagia without success [19].

We present a case series of 10 pediatric patients suffering from chronic upper respiratory symptoms, which were usually made worse by activity. These patients were found to only have the diagnosis of LTH, diagnosed on flexible bronchoscopy by a pulmonologist, and were refractory to medical treatment. With consideration of all time points, we saw significant symptom resolution in most of our patients due to lingual tonsillectomy.

Our preliminary case series may be indicative of the role of LTH in a variety of chronic upper respiratory issues. However, our study is not without limitations, including small sample size, the retrospective nature of the study, and lack of standardization of the pre-operative and post-operative work-up (the only consistent factor was that all patients underwent pre-operative flexible pulmonary bronchoscopy). We recommend future studies be conducted with a larger patient population in order to analyze the role of lingual tonsillectomy in refractory upper airway symptoms (such as exercise-induced shortness of breath, and chronic cough/throat-clearing/globus sensation). In conclusion, lingual tonsillectomy significantly resolved chronic upper respiratory symptoms, usually related to activity, that were nonresponsive to other medical treatments. These symptoms include but are not limited to cough, throat clearing, and shortness of breath. Our study indicates that hypertrophic lingual tonsils should be considered as a contributing factor to these types of symptoms, but further studies are needed to elucidate its role.

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