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The safety and efficacy of pediatric lingual tonsillectomy

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ABSTRACT

Objective: Lingual tonsillar hypertrophy is recognized as a cause of persistent obstructive sleep apnea (OSA) after adenotonsillectomy in children. However, little has been reported regarding the complications, postoperative course and effectiveness of lingual tonsillectomy (LT). Our objective was to review the safety and effectiveness of LT in children.
Methods: Retrospective review of children undergoing LT from January 2009 to December 2015 at a tertiary children's hospital. Complications, postoperative course and polysomnographic (PSG) outcomes were recorded for all patients.
Results: We identified 92 children (mean age = 8.6 years, 50% female) who underwent LT; 43.5% had a syndromic diagnosis. The most common complications were emergency department presentation for bleeding (4.4%) and poor oral intake (3.3%). The readmission rate was 4.4% including 2 children (2.2%) who required operative control of hemorrhage. No children required unplanned reintubation or ICU admission. In children with PSG data (n = 18), the median apnea-hypopnea index (AHI) decreased from 8.5 to 3.8 events/hour ($p = 0.022$) and the median obstructive AHI (oAHI) decreased from 8.3 to 3.1 events/hour ($p = 0.021$). In addition, the oxygen saturation nadir increased from 83.8% to 89.0% ($p = 0.0007$). After surgery the percentage of patients with oAHI<5 events/hour increased from 27.8% to 61.1% ($p = 0.08$).
Conclusions: Readmission and bleeding rates after lingual tonsillectomy in children were similar to that seen with tonsillectomy. Polysomnographic data showed that lingual tonsillectomy resulted in a significant reduction of both AHI and oAHI with a postoperative oAHI <5 achieved in 61% of patients.

1. Introduction

The lingual tonsils are composed of lymphoid tissue situated at the base of the tongue; they constitute one component of Waldeyer's ring, which was first described by Vesalius in 1543 [1]. Lingual tonsil hypertrophy was further described four hundred years later by Elia [2] in patients with lingual tonsillitis following adenotonsillectomy. Two additional reports later characterized the lingual tonsils as a possible site of acute upper airway obstruction and as a source of tonsillitis in patients who had previously undergone adenotonsillectomy [3,4]. Eventually lingual tonsils were identified as a possible anatomical cause for obstructive sleep apnea (OSA) and a trial of lingual tonsillectomy was carried out in adults in 2003 [5].

In children, removal of the tonsils and adenoids (T&A) is the standard first line surgical therapy for OSA. However, it has become evident that up to 40% of children will have persistent OSA following T&A [6]. Causes of persistent OSA following T&A include nasal deformity, inferior turbinate hypertrophy, regrowth of adenoids, retro-palatal collapse, oropharyngeal collapse, glossoptosis, laryngomalacia and lingual tonsillar hypertrophy. Lingual tonsillectomy was first reported in a child with persistent OSA in 2006 by Kluszynski [7] and colleagues. Since that time, lingual tonsillectomy has become one of the most commonly described procedures

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employed for children with persistent OSA [8]. While several other small studies have evaluated the effectiveness of lingual tonsillectomy, none have described associated complications [9–12]. In light of these findings, the objective of our study was to describe the short-term complications as well as the effectiveness of lingual tonsillectomy in children.

2. Materials and methods

A retrospective chart review was performed identifying patients who underwent lingual tonsillectomy, for any indication, between January 1st, 2009 and December 31st, 2015. All patients <18 years of age were included for review. Patients were excluded if they underwent concurrent upper airway surgery including tonsillectomy, adenoidectomy, nasal procedures, midline posterior glossectomy or laryngeal/tracheal procedures. Patients with follow-up less than 3 weeks were considered to have inadequate short-term follow-up and were also excluded. Institutional review board approval (IRB) was obtained prior to data collection (CCHMC IRB: 2015–2260).

Prior to surgery, patients underwent complete office physical exam including flexible fiber optic laryngoscopy when tolerated. In patients with OSA, drug induced sleep endoscopy (DISE) was performed at the surgeon's discretion. All patients with persistent OSA (AHI>1) were offered medical therapy (eg. nasal steroids) and those with moderate or severe OSA were offered CPAP prior to surgery. Lingual tonsillectomy was offered in children with persistent OSA, clinical symptoms and lingual tonsillar hypertrophy. Children were evaluated for lingual tonsillitis when symptoms of recurrent or chronic pharyngitis persisted despite T&A. Surgery was offered when flexible endoscopy demonstrated lingual hypertrophy.

Lingual tonsillectomy was performed under general anesthesia, utilizing orotracheal or nasotracheal intubation. Surgery was performed via laryngoscopic direct visualization with a Lindholm scope or through the endoscopic approach described by Maturo et al. [13,14]. Both suction cautery and coblation were utilized for removal of lingual tonsils. Patient electronic medical records (EMR) were reviewed for surgical technique, hospital stay, immediate post-operative complications, emergency department (ED) presentation, short-term complications, need for revision surgery and polysomnographic data. All patients included had clinic or telephone follow-up 3 or more weeks after surgery with specific questioning about bleeding, emergency room presentation and other postoperative complications.

2.1. Polysomnography group

A subset of patients was identified who underwent lingual tonsillectomy for persistent OSA and had available preoperative and postoperative sleep studies. Results of overnight polysomnography (PSG) were reviewed via the electronic medical record system. Polysomnography data collected included sleep parameters, apnea hypopnea index (AHI), obstructive apnea hypopnea index (oAHI), Oxygen saturation (O_2) nadir, and % time with CO_2 > 50 mm Hg. OSA was categorized by severity, with an oAHI of 1–5 defined as mild, oAHI of 5–10 as moderate and oAHI of >10 as severe OSA.

2.2. Statistical analysis

Demographic data was summarized for categorical and continuous measures. Means and ranges are presented for continuous normalized data. Medians are presented for non-normal data. Categorical variables are presented as percentages. Continuous data was compared between groups using Wilcoxon rank sums test for non-normal data and Fisher's exact test for categorical measures. Categorical data was compared among groups using chi-square analysis and Fisher's exact tests as appropriate. Pre and post-operative changes in sleep data within each subject were tested using Wilcoxon signed rank or McNemar's test (for paired analysis). All analyses will be performed using SAS® for Windows (SAS Institute Inc., Cary, NC USA).

3. Results

3.1. Demographics

189 patients underwent lingual tonsillectomy during the study period. Of these children, 99 had lingual tonsillectomy alone; 92 of these patients had adequate follow-up for inclusion. These children had a median age of 8.6 years (mean age = 8.4 years, range 2.1–17.9 years) at the time of surgery; 50% were female, and 87% were white. Comorbid diagnoses were recorded in 43.5% of these children; Down syndrome (DS) was identified as the most common comorbidity (28.3%). [Table 1].

3.2. Surgery and hospital course

The most common indication for surgery was OSA (n = 57,62.0%) followed by recurrent lingual tonsillitis (n = 32, 34.8%). With respect to surgical technique, 51 (55.4%) surgeries were performed using coblation lingual tonsillectomy, while suction cautery was used in 41 (44.6%) cases [Table 2]. Following surgery, the median length of stay (LOS) was 1 day (mean LOS = 0.8 [0–4] days) and no patients experienced unplanned intubation or unplanned intensive care unit (ICU) admission. Median follow-up for all patients was 5.9 months (mean = 7.9 [0.6–71.6] months). [Table 2].

3.3. Postoperative complications

Presentation to the ED was recorded for 9 (9.8%) children after surgery. During ED evaluation patients complained of bleeding (n = 4, 4.4%), voice change (n = 4, 4.4%), and decreased oral intake (n = 3, 3.3%). Readmission was recorded in 4 (4.4%) children; bleeding and decreased oral intake were the most common reasons for readmissions. The overall bleeding rate for lingual tonsillectomy was 4.4%. Bleeding was further categorized as patient reported without evidence of bleeding on physical examination (n = 1, 1.1%), confirmed bleeding treated with observation only (n = 1, 1.1%), and confirmed bleeding with operative intervention (n = 2, 2.2%). There

Table 1
Demographic characteristics of patients undergoing lingual tonsillectomy.

Patient characteristic	Value
No. of Patients	92
Age, yr, median, (mean) [range]	8.6, (8.4) [2.1–17.9]
Gender (% female)	50.0%
BMI at Surgery %, median, (mean) [range]	71.7, (91) [1.0–99.7]
Race, n (%)	
White	80 (87.0%)
Black	2 (2.1%)
Other/Unknown	10 (10.9%)
Syndrome Diagnosis, n (%)	40 (43.5%)
Down Syndrome	26 (28.3%)
Charge Syndrome	4 (4.4%)
Other Syndromes	8 (8.7%)
Other Comorbid Conditions, n (%)	
Tracheostomy Dependence	10 (10.9%)
Neurologic Malformations	2 (2.1%)
Other Syndromes	8 (8.7%)

Table 2
Surgery and hospital course.

Patient characteristic	Value
No. of patients	92
Surgical Indication, n (%)	
Obstructive Sleep Apnea (OSA)	57 (62%)
Recurrent Tonsillitis	32 (34.8%)
Other	3 (3.3%)
Surgical Technique, n (%)	
Coblation	51 (55.4%)
Suction Cautery	41 (44.6%)
Hospital Course, n (%)	
Unplanned ICU Admission	0 (0.0%)
Re-Intubation	0 (0.0%)
Length of Stay, days, median (mean) [Range]	1.0 (0.8) [0–4]
Follow-up, median (mean), [range]	5.3, (7.9) [0.6–71.6]

was no significant difference in bleeding or complication rates when comparing the surgical techniques used or when comparing children with syndromes to typical patients [Tables 3 and 4].

3.4. Sleep outcomes

Of the 57 children who underwent lingual tonsillectomy for the treatment of OSA 30 patients had an available preoperative sleep study with median and mean oAHI of 8.3 and 11.9 respectively. 18 (31.6%) had complete preoperative and postoperative PSG data. Of the patients with available preoperative and postoperative sleep study data, 13 (72.2%) had a syndrome; DS was the most common diagnosis (11/13). The median time from surgery until post-operative sleep study was 7.6 months (mean = 8.5 months) in this group. Following surgery, the median post-operative change in AHI was 5.3 events/hour (mean = 6.9 [0.6–71.6]). AHI decreased from a median (mean) value of 8.5 (12.8) to 3.8 (5.9) events/hour after surgery ($p = 0.022$); similarly, the median (mean) oAHI decreased from 8.3 (12.4) to 3.1 (5.5) events/hour after surgery ($p = 0.021$) [Table 5]. In addition, the median (mean) oxygen saturation nadir was increased from 83.8% (81.2%) to 89.0% (87.8%) ($p = 0.0007$). Body mass index (BMI) percentile, sleep efficiency, percentage of rapid eye movement (REM) sleep, arousal index, OSA severity, and %CO2 > 50 mm Hg did not change significantly following surgery. After lingual tonsillectomy, 61.1% (11/18) of the children were found to have an oAHI <5 events/hour compared to 27.8% (5/18) who had an oAHI < 5 prior to surgery; one child (5.6%) had a post-operative AHI of <1.0 event/hour ($p = 0.032$) [Table 5]. Of note, 3 children with an oAHI of <5 preoperatively had a post-operative oAHI of >5 postop.

4. Discussion

Although pediatric lingual tonsillectomy has previously been described, this study is the first to examine both short-term complications and effectiveness [9–12]. Our findings suggest that lingual tonsillectomy is a safe procedure with low rates of ED presentation, re-admission, bleeding and need for control of hemorrhage. In addition, available PSG data demonstrates that lingual tonsillectomy can significantly improve the AHI, oAHI, and oxygen saturation nadir.

For the past six decades clinicians have sought an effective treatment to address hypertrophy of the lingual tonsils. In the 1950s, radiation therapy was recommended as the primary means to treat lingual tonsillar hypertrophy [2]. At that time, surgical removal of the lingual tonsils was considered a dangerous procedure with the possibility for significant acute and chronic complications [2]. Injury to adjacent blood vessels and subsequent severe bleeding were of particular concern [15]. A wide variety of surgical techniques, including suction bovie cautery, CO2 laser, microdebrider, and coblation, were then developed in order to more safely remove the lingual tonsils [3,7,14,16]. In recent years, suction bovie cautery and coblation have emerged as the most widely utilized techniques for lingual tonsillectomy; both of these techniques were assessed in our study.

Recent literature describes lingual tonsillectomy as "safe" without specific evaluation or enumeration of the likelihood of complications [1,14,16]. In our study 9.8% of children presented to the ED following lingual tonsillectomy and 4.4% of patients required readmission. This readmission rate is comparable to the readmission rate (7.8%) published recently by Mahant et al. [17] for 139,715 children undergoing adenotonsillectomy in the United States. Our patients had an overall bleeding rate of 4.4%, which is similar to the 3.3% bleeding rate reported in Krishna and colleague's meta-analysis of tonsillectomy complications [18].

There is general concern that patients with a syndromic diagnosis are at increased risk for post-surgical complications. In children with Down syndrome, studies have shown an increased risk of bleeding and postoperative respiratory complications following adenotonsillecotmy [19,20]. However, our study demonstrates no evidence of increased bleeding or complication rates in children with syndromes. In addition, in our cohort, complications were not influenced by the surgical method used for lingual tonsil removal.

Our review of 92 patients also did not identify any children who required ICU transfer or reintubation. Hospital stay was relatively short, ranging from 0 to 4 days, with a median and mean stay of 1 day and 0.8 days, respectively.

The most common reason for lingual tonsillectomy in our study was persistent OSA after T&A. A recent systematic review established that 34% of children will have persistent moderate to severe OSA following T&A [7]. Persistent OSA is especially common in children with craniofacial abnormalities, obesity and syndromic diagnosis [21–24]. Children with Down syndrome (DS) are noted to have a 48% chance of persistent OSA with the lingual tonsils

Table 3
Postoperative complications by surgical technique.

Outcome/Complication	All Patients	Coblation	Cautery	P Value
No. of patients (%)	92	51 (55.4%)	41 (44.6%)	–
Complications, n (%)				
Emergency Department Presentation	9 (9.8%)	6 (11.8%)	3 (7.3%)	0.73
Readmission	4 (4.4%)	4 (7.8%)	0	0.13
Dysphagia/Decreased Oral Intake	3 (3.3%)	1 (2%)	2 (4.9%)	0.58
Voice Change	4 (4.4%)	3 (5.9%)	1 (2.4%)	0.63
Bleeding, n (%)				
Any Bleeding	4 (4.4%)	4 (7.8%)	0	0.13
Reported bleeding	1 (1.1%)	1 (1.1%)	0	–
Confirmed Bleeding: Observation	1 (1.1%)	1 (1.1%)	0	–
Confirmed Bleeding: Operative	2 (2.2%)	2 (2.2%)	0	–

Table 4
Hospital course and postoperative complications by syndrome diagnosis.

Outcome/Complication	All Patients	Syndromic	Non-syndromic	P Value
No. of patients (%)	92	40 (43.5%)	52 (56.5%)	–
Complications, n (%)				
Emergency Department Presentation	9 (9.8%)	3 (7.5%)	6 (11.5%)	0.73
Readmission	4 (4.4%)	2 (5%)	2 (3.9%)	1.0
Dysphagia/Decreased Oral Intake	3 (3.3%)	0	3 (5.8%)	0.25
Voice Change	4 (4.4%)	2 (5%)	2 (3.9%)	1.0
Bleeding, n (%)				
Any Bleeding	4 (4.4%)	3 (7.5%)	1 (1.9%)	0.32
Reported bleeding	1 (1.1%)	1 (2.5%)	0	–
Confirmed Bleeding: Observation	1 (1.1%)	0	1 (1.9%)	–
Confirmed Bleeding: Operative	2 (1.1%)	2 (5.0%)	0	–

Table 5
Sleep study findings for obstructive sleep apnea (OSA) patients (N = 18).

	Preoperative	Postoperative	P-Value
BMI %, median [range]	88.3 [1–100]	88.9 [2.8–100]	0.81
Sleep Efficiency median [range]	82.8 [21.4–96.5]	87.4 [66.7–96.2]	0.60
% REM median [range]	20.6 [0–27.6]	19.8 [2.3–63.3]	0.82
Arousal Index median [range]	14.6 [4.2–30.9]	12.5 [4.4–23.9]	0.15
Apnea Hypopnea Index (AHI), median (mean) [range]	8.5 (12.8) [1.5–43.7]	3.8 (5.9) [0.5–19.1]	**0.022**
Obstructive AHI (oAHI), median (mean) [range]	8.3 (12.4) [1.4–43.4]	3.1 (5.5) [0.1–18.2]	**0.021**
Difference in AHI after surgery, median (mean) [range]		5.3 (6.9)[13.9–42.5]	
Difference in oAHI after surgery, median (mean) [range]		5.6 (6.9) [13.6–42.2]	
Severity of Obstructive Sleep Apnea, n (%)			
None	0	1 (5.6%)	0.61
Mild	5 (27.8%)	10 (55.6%)	
Moderate	6 (33.3%)	4 (22.2%)	
Severe	7 (38.9%)	3 (16.7%)	
%CO_2 > 50 mm Hg, n (%)	8.1 [0–91]	2.8 [0–96.3]	0.56
Oxygen saturation nadir, median (mean) [range]	83.8 (81.2)[51.9–94.8]	89 (87.8) [72.7–93.9]	**0.0007**
AHI <1 events/hour, n (%)	0	1 (5.6%)	0.32
AHI <5 events/hour, n (%)	3 (16.7%)	11 (61.1%)	**0.01**
oAHI <1 events/hour, n (%)	0	1 (5.6%)	0.32
oAHI <5 events/hour, n (%)	5 (27.8%)[a]	11 (61.1%)	0.08

BMI = body mass index, REM = rapid eye movement, AHI = apnea hypopnea index, oAHOI = obstructive AHI, CO2 = carbon dioxide, Hg = mercury.
The P-Values bolded are statistically significant.
[a] NOTE THAT 3 OF THE 5 WHO HAD OAHI <5 PRE HAD OAHI>5 POST.

identified as a source of persistent airway obstruction in 30% of these children [25,26]. Children with DS were common in our series and represented 28.3% of our cohort.

Manickam et al. [8] recently published a systematic review of obstruction site identification in children with persistent OSA. This review identified 4 small studies evaluating lingual tonsillectomy with a combined sample size of 141 children. The authors reported OSA resolution rates ranging from 57% to 88%. Several factors, however, limit the broad applicability of the included studies. The studies by Truong et al. [9] and Chan et al. [11] included patients undergoing concurrent procedures in addition to LT. The series (n = 16) by Abdel-Aziz et al. [12] demonstrated a reduction of mean AHI to < 5 events/hour, with no mention of statistical significance. The article by Lin et al. [10] studied 26 patients and noted that lingual tonsillectomy resulted in significant improvement in both the mean respiratory distress index (RDI) from 14.7 to 8.1 events/hour (P = 0.02) and obstructive apnea index from 18.1 to 2.2 events/hour (p = 0.038). However, none of the included studies addressed short-term complications, ED presentation, need for re-admission or bleeding rate.

Similar to previous published studies, our research demonstrates a significant improvement in both AHI and oAHI with lingual tonsillectomy. For comparison it is important to note that most previous studies reported means, while we are reporting medians. The choice to report a median with a range was based on the distribution of the data, which was fairly skewed and had large measures of variability (large standard deviations). Based on the non-normal data distribution, the medians were a more appropriate statistical measure. When directly comparing means our reductions of AHI and oAHI are similar to previous publications [9–11].

However, unlike many of these previous studies, all of the children included in our study underwent isolated lingual tonsillectomy.

Our surgical success results are in keeping with rates reported in the literature with 61.1% of the children in our study successfully treated (defined as a postoperative oAHI<5 events/hour). These findings suggest that lingual tonsillectomy can effectively reduce oAHI for a significant proportion of children. It should be noted that 3 patients with mild OSA had worsened oAHI on post-operative PSG. In these patients, lingual tonsillar hypertrophy may not be the only reason for persistent obstruction. It is possible that removal of the lingual tonsils may result in worsened obstruction secondary to glossoptosis. In addition, median time to sleep study was 7.6 months in our sleep outcomes cohort. During this time changes in body habitus may have contributed to worsening of OSA.

The present study is not without limitations. The data was collected in a non-randomized, retrospective manner without standardized protocols. While recurrent pharyngitis was a common indication for surgery, follow-up in these patients was limited to 1 or 2 visits postoperatively. We cannot, therefore, make any statements about the effectiveness of lingual tonsillectomy in the setting of recurrent pharyngitis. Our sleep outcome findings are limited to the 31.6% of patients with OSA who had complete polysomnography data. This is in part due to a lack of a standardized polysomnography protocol and the tertiary nature of our facility. However, this may reflect a selection bias. To address these issues our institution has developed an Upper Airway Center to standardize evaluation, treatment and follow-up in complicated patients with OSA. Our sleep outcome data is also biased given that a majority of patients (72.2%) with a postoperative sleep study had a syndromic diagnosis. In the future prospective evaluations of lingual tonsillectomy outcomes with long-term follow-up are recommended to confirm our findings regarding the safety and effectiveness of lingual tonsillectomy.

5. Conclusions

This study demonstrates that lingual tonsillectomy is safe with comparable complication rates to adenotonsillectomy. In patients with adequate sleep follow-up, lingual tonsillectomy significantly reduces both AHI and oAHI, with oAHI <5 achieved in 61.1% of patients.

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Conflict of interest

None.

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