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Lingual tonsillectomy in children with Down syndrome: Is it safe?

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ABSTRACT

Objective: Evaluate peri-operative course and morbidity in children with Down syndrome (DS) who underwent a lingual tonsillectomy (LT) for residual obstructive sleep apnea (rOSA).
Methods: Retrospective case series for children with DS who underwent LT for rOSA from April 2011 to July 2016. Our primary outcomes were length of stay, readmission and complications. Surgical effectiveness was evaluated by change in the obstructive apnea-hypopnea-index(OAHI) and oxygen saturation nadir.
Results: Thirty-nine patients underwent LT. The mean length of stay was 1.3 days with n = 21(72%) staying one night. One subject (2.6%) had a post-operative bleed that did not require operative intervention. No other major complications occurred. In terms of effectiveness of surgery, twenty–nine children had sufficient data for inclusion. Median OAHI did not appreciably change ($p = 0.07$) from before surgery. Five subjects (17%) were cured of OSA (OAHI < 2/hour) and a mix of improvement and worsening was identified. The lowest oxygen saturation improved from 78% (SD = 7) before surgery to 82% (SD = 6) after surgery ($p = 0.003$).
Conclusion: LT has a favorable post-operative course but its effectiveness at curing rOSA in the DS population has not been established/proven. Further research is indicated to determine optimal surgical management for DS children with LTH.
Level of evidence: 4.

1. Introduction

Obstructive sleep apnea (OSA) is common in children with Down syndrome (DS) with a prevalence of up to 80% [1]. Macroglossia, midface hypoplasia, hypotonia, lymphoid hyperplasia and body habitus predispose these children to OSA [2,3]. Unfortunately, adenotonsillectomy (T&A) is rarely curative [4–7]. Children with residual OSA (rOSA) are often treated with continuous positive airway pressure (CPAP) [8]. The tolerance of these devices in children with DS is limited, even with significant attempts to improve tolerance and slow device introduction [9,10]. The sequelae of untreated OSA include pulmonary hypertension, cardiovascular complications, failure to thrive and impaired cognition [11–14]. Treatment of rOSA is important for any child.

The surgical treatment options for rOSA in children include pharyngeal surgeries (uvulopalatopharyngoplasty) [15,16] hypopharyngeal procedures such as lingual tonsillectomy(LT) and tongue base reduction [3,17] and laryngeal (supraglottoplasty, epiglottopexy) [18,19]. Recently drug induced sleep endoscopy (DISE) has increasingly been utilized to facilitate surgical planning [20,21]. A common site of obstruction in patients with rOSA following a T&A is the hypopharynx [21,22]. For those children with lingual tonsil hypertrophy (LTH), a LT is a surgical option [23,24]. Its effectiveness for children is fair, depending on the definition of success. A recent meta-analysis identified a mean improvement in AHI of 9 points with 17% cure rate (post-operative AHI < 1) [25]. The success rate was 51% (post-operative AHI of < 5) but the proportion of subject starting with an AHI < 5 is unclear from the studies contributing to the meta-analysis. Recognizing that improvement in AHI with LT is at best fair and may be worse for the DS population, the procedure's morbidity takes center stage. Previous assessments of the morbidity associated with pediatric LT include a variety of indications in addition to OSA including lingual tonsillitis, which comprised nearly 35% of the studied subjects [26]. The morbidity associated with hypopharyngeal surgery in the rSOA population as well as the DS population may be different.

Our objective is to assess LT peri-operative course to provide clinicians with data that will facilitate shared decision making with the family when they are deciding whether to proceed with an operation that has a 50% chance of improving the OAHI to less than 5 events/hour.

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2. Methods

2.1. Study subjects

This retrospective case series study analyzed DS children who underwent a LT from April, 2011 to July 2016. Subjects having additional surgeries of the nasal passage, palate or epiglottis were included; however subjects having concurrent base of tongue reductions were excluded. Subjects with rOSA (OAHI ≥ 2.0) were included in the treatment effectiveness analyses. No patients were lost to follow up however some failed to tolerate post-operative polysomnography (PSG) [n = 8]. The Colorado Multiple Institutional Review Board approved the project.

2.2. Treatment protocol

Patients underwent LT by seven surgeons for rOSA and LTH. The surgical technique has previously been described [23]. Briefly, patients underwent nasotracheal intubation. Access to the oral cavity was facilitated with either a Jennings mouth gag or dental bite block. Visualization of the hypopharynx was achieved with either a sweetheart retractor or Parsons laryngoscope and a telescope. All surgeons used the Coblator(TM) device to perform the procedure. All patients were admitted post-operatively.

2.3. Data collection

Potential cases were identified based on billing code (CPT 42870) and confirmed by reviewing the operative report. Electronic medical records were reviewed for inclusion and exclusion criteria. Demographic data, morbidity and effectiveness data was collected using RedCap.

2.4. Outcomes

Our primary outcomes were length of stay, readmission and rate of complications. Events were considered post-operative complications if they occurred within the 3 weeks after surgery (Table 1). Descriptive statistics were calculated for the post-operative complications and length of stay for all patients that underwent LT. Phone notes and electronic message log system were queried to identify potential post-operative morbidity not identified in readmission. To understand risk factors for increased length of stay, we tested for differences in length of stay by OSA category and the performance of multiple procedures. PSGs were scored by an experienced sleep and electroencephalography-registered technologist. For surgical effectiveness, we evaluated change OAHI and change in lowest oxygen saturation.

Table 1
Minor and major post-operative complications after lingual tonsillectomy.

Complication	n (%)
Major Complications	
Obstruction requiring jaw thrust, positive pressure, or reintubation	–
Respiratory depression requiring intervention, (i.e. naloxone)	–
Unplanned PICU admission	–
Post-obstructive pulmonary edema	–
High flow 0_2 (100% by face mask or non-rebreather)	–
Immediate post-op hemorrhage (< 24 h post-op)	–
Post-op hemorrhage (> 24 h post-op) requiring surgery	–
Minor Complications	
Minor obstruction requiring suction, or nasal airway	–
Minor obstruction requiring oxygen only	11 (28%)
Repositioning	–
Oxygen desaturation < 80%	–
Oxygen saturations persistently < 90%	–
Post-op vomiting	1 (3%)
Bleeding not requiring OR, spontaneously resolved	1 (3%)
Dehydration	3 (8%)
Other	–

2.5. Data analysis

Post-operative stay and complications data was collected including length of stay, rate and character of minor and major post-operative complications. Events were considered post-operative complications if they occurred within the 3 weeks after surgery. Descriptive statistics were calculated for the post-operative complications and length of stay for all patients that underwent LT.

The treatment effect of LT was tested for change in the median OAHI before and after surgery with the Wilcoxon signed-rank test. Change in lowest 02 saturation was tested with paired *t*-test. To further describe and define the sleep apnea severity before and after LT, subjects were categorized into no OSA (OAHI < 2), mild OSA (OAHI 2 - < 5), moderate OSA (OAHI 5- < 10) and severe OSA (OAHI ≥10) both pre and post LT [27]. As exploratory analysis, we tested whether subgroups including those with BMI percentile < 95% or OAHI < 10 predicted a more favorable outcome.

3. Results

Thirty nine patients underwent LT during the study timeframe for inclusion in the length of stay and complication assessment. Baseline characteristics (Table 2) show a slight male (59%) and Caucasian (56%) preponderance. Pre-operatively, 90% (35/39) children had a LTH grade of either III (where the lingual tonsil either fills the vallecula) or IV (the lingual tonsil obscures the epiglottis) [28]. Additional therapeutic procedures in addition to LT included revision adenoidectomy (n = 9), inferior turbinate reduction (n = 9), uvulopalatopharyngoplasty (n = 6), epiglottopexy (n = 3) and revision tonsillectomy (n = 1).

Length of stay and post-operative complications were available on n = 39 subjects. Overall, mean length of stay was 1.3 days with n = 29 (74%) staying one night post op and n = 9 (23%) staying two nights post op and one subject (3%) staying 3 nights. There were only 3 patients admitted electively to the PICU. Two had severe OSA and required non-invasive ventilation and one had severe developmental delay in addition to Down syndrome and was admitted at the request of anesthesia who did not want to extubate the child initially. All three of these children had an uneventful night in the PICU and were discharged on post-operative day #1. Overall, sixteen subjects (41%) had minor complications. No major complications occurred in the case series. Most minor complications were periodic desaturations but not consistently

Table 2
Characteristics of subjects with DS undergoing lingual tonsillectomy.

Parameter	All Subjects
	n = 39
Child's Age; mean ± SD years	11 ± 4 [Range 3.7–17.9]
Child's Sex; n (%) Female	16 (41%)
Lingual Tonsil Grade	
II	4 (10%)
III	20 (51%)
IV	15 (38%)
Lowest 02 Sat	78 ± 7
Saturation < 90%, percent TST	6 [1–23]
OAHI, median TST	10 [6–27]
OAHI, median REM sleep[a]	21 [11–44]
CAHI, mean TST[b]	0.2 [0–1.3]

Data presented as n (%), mean ± SD and median [IQR].
Standard Deviation (SD), Polysomnogram (PSG), end tidal carbon dioxide (ET CO2), total sleep time (TST), obstructive apnea hypopnea index (OAHI), rapid eye movement (REM), central apnea index (CAI), interquartile range (IQR).
[a] Unavailable 8 subjects.
[b] Unavailable on 16 subjects.