Oropharyngeal Stenosis

A Complication of Multilevel, Single-Stage Upper Airway Surgery in Children

Jeremy D. Prager, MD; Brandon S. Hopkins, MD; Evan J. Propst, MD; Sally R. Shott, MD; Robin T. Cotton, MD

Objectives: To describe oropharyngeal stenosis (OPS), a potential complication of multilevel, single-stage upper airway surgery involving lingual tonsillectomy in children, and to discuss the manner in which OPS may be managed successfully.

Design: Case series with an average follow-up of 12 months.

Setting: Tertiary care children’s hospital.

Patients: Medical charts were reviewed for 104 patients who underwent lingual tonsillectomy over a 30-month period from January 1, 2007, to June 30, 2009.

Intervention: Multilevel, single-stage upper airway surgery, including lingual tonsillectomy.

Main Outcome Measure: Development of OPS noted during office or intraoperative examination.

Results: Forty-nine of 104 patients underwent multilevel, single-stage upper airway procedures that included lingual tonsillectomy. Four of these 49 patients developed OPS, for a complication rate of 8.2%. Three patients required pharyngoplasty (scar release, debulking of fibrotic tissue, and reorientation of the scar) and triamcinolone injections in the operating room. A fourth patient underwent simple scar release in the operating room. No patient who underwent lingual tonsillectomy alone or in combination with an additional procedure at the same level of the upper airway developed OPS.

Conclusions: Oropharyngeal stenosis is a potential complication of multilevel, single-stage upper airway surgery involving lingual tonsillectomy in children. Although there is pressure to perform multilevel procedures that address each site of upper airway obstruction in 1 sitting, this case series suggests the need for a more conservative, staged approach if lingual tonsillectomy is planned.


Oropharyngeal stenosis (OPS) is a narrowing of the upper aerodigestive tract in the region of the soft palate, lateral pharyngeal walls, and tongue base. The narrowing may appear as a cicatrical scar that is visible in the oropharynx and caused by adhesion of the anterior pillars and inferior tonsillar fossae to the tongue base, which accordingly narrows the oropharyngeal aperture. Symptoms of dyspnea on exertion, dysphagia, poor weight gain, sleep-disordered breathing, and obstructive sleep apnea (OSA) may result from OPS. Some patients with OPS may be asymptomatic.

In the past, OPS has been described as a rare complication of tonsillectomy with or without adenoidectomy (T±A). Deep dissection of the lower tonsillar pole and removal of adjacent lingual tonsil tissue may cause OPS. Excessive cautery and postoperative bleeding have been associated with nasopharyngeal stenosis and OPS. Other potential pathogenetic causes include oropharyngeal infection, most notably syphilis, although other infectious causes have been identified.

With increased awareness of persistent airway obstruction despite previous T±A, other upper airway structures are being surgically addressed with increasing frequency. These surgical solutions include methods of nasal airway improvement, soft palate reduction, lingual tonsillectomy, and procedures to reduce the tongue base. Many new techniques and devices have been used in these procedures. Multilevel procedures are those that address structures in several upper airway regions: the nasal airway, nasopharynx, soft palate, palatine tonsils and their pillars, lingual tonsils, and tongue base. According to this definition, adenotonsillectomy is a multilevel, single-stage procedure. The trend has been to treat multilevel upper airway obstruction with multilevel procedures performed simulta-
obtained for this retrospective case series. Institutional review board approval was
secured. Data were collected in a deidentified, pass-word-secured location. The review
included operative reports and clinical notes from before, during, and after this pe-
riod. Patients who underwent multilevel, single-stage procedures, either alone or in
combination with procedures at the same level of the upper airway (0 of 56 patients).
The clinical courses of these 4 patients are presented in Table 2 in order of their pre-
sentation. The average age of these patients at the time of their multilevel, single-stage procedure was 4.5 years (age range, 3 years 4 months to 5 years 8 months). Each pa-
tient required intervention in the operating room. Follow-up for these patients averaged 1 year 5 months (range, 11-24 months).

Each of the 4 patients with OPS experienced clinical setbacks as a result of his or her OPS. Patient 1 was diag-
osed as having OSA by polysomnography after his OPS was noted and required continuous positive airway pres-
sure for several months while the OPS was being treated. He has since been weaned from this support. Patient 2 had worsening of his OSA on polysomnography because of OPS and required oxygen supplementation, which was eventually discontinued during treatment of his OPS. He is not currently being treated with continuous positive airway pressure and has yet to undergo another poly-
somnography. Patient 3, a girl, underwent an examination under anesthesia and unanticipated intraoperative scar division. Patient 4 experienced delayed decannula-
tion (6 months) as she underwent multiple additional procedures to improve her OPS. Patient 2, whose course is representative, is described herein in greater detail.

Patient 2 was a nearly 3½-year-old boy with a history of trisomy 21. He was diagnosed as having OSA by polysomnography after his OPS was noted and required continuous positive airway pressure for several months while the OPS was being treated. He has since been weaned from this support. Patient 2 had worsening of his OSA on polysomnography because of OPS and required oxygen supplementation, which was eventually discontinued during treatment of his OPS. He is not currently being treated with continuous positive airway pressure and has yet to undergo another poly-
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METHODS

After the discovery of several patients with OPS who had undergone multilevel, single-stage procedures that included lingual tonsillectomy, medical records were reviewed for those pa-
tients who had undergone lingual tonsillectomy from January 1, 2007, to June 30, 2009, at Cincinnati Children’s Hospital Medi-
cal Center, Cincinnati, Ohio. The review included operative re-
ports and clinical notes from before, during, and after this pe-
riod. Patients who underwent multilevel, single-stage procedures, including lingual tonsillectomy, were noted and closely re-
viewed as a separate population. The treatment of patients with
OPS was examined. Data were collected in a deidentified, pass-
word-secured location. Institutional review board approval was
obtained for this retrospective case series.

RESULTS

A total of 104 individuals underwent 105 separate lingual tonsillectomy procedures during the specified period (1 pa-
tient underwent lingual tonsillectomy alone and again as part of a multilevel, single-stage procedure at a later date). The most common indication was upper airway obstruc-
tion, followed by recurrent pharyngitis. Fifty-six patients underwent single-level, single-stage surgery: lingual tonsillectomy alone or lingual tonsillectomy in combination with other procedures at the same level of the upper air-
way (radiofrequency ablation of the tongue base or poste-
rrior midline glossectomy). Of these 56 patients, 35 under-
went follow-up examinations, none of which revealed OPS.

Forty-nine patients underwent multilevel, single-stage surgical procedures that included lingual tonsillectomy. No patients underwent more than 1 multilevel procedure. Twenty-four of the patients (49%) were fe-
male, and 25 were male. No patients were noted to have OPS at their first examination in the operating room or clinic. The average age at the time of procedure was 10 years (age range, 1-21 years; SD, 5 years 2 months). The average duration of follow-up was approximately 12 months (range, 0-32 months; SD, 10 months). Twelve patients (24%) did not receive follow-up. The distribu-
tion of procedures, broken down by multilevel, single-stage surgery vs single level, single-stage surgery and pres-
ence or absence of OPS, is presented in Table 1.

Four patients who underwent multilevel, single-stage surgery involving lingual tonsillectomy developed OPS, for a complication rate of 8.2%. This compares with the 0% incidence of OPS in the population of patients who underwent lingual tonsillectomy either alone or in combi-
nation with procedures at the same level of the upper airway (0 of 56 patients). Twelve patients (24%) did not receive follow-up. The distribution of procedures, broken down by multilevel, single-stage surgery vs single level, single-stage surgery and pres-
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somnography. Patient 3, a girl, underwent an examination under anesthesia and unanticipated intraoperative scar division. Patient 4 experienced delayed decannula-
tion (6 months) as she underwent multiple additional procedures to improve her OPS. Patient 2, whose course is representative, is described herein in greater detail.

Patient 2 was a nearly 3½-year-old boy with a history of trisomy 21. He was diagnosed as having OSA and
underwent an uncomplicated adenotonsillectomy, which failed to resolve the problem. Magnetic resonance imaging demon-
strated lingual tonsil hypertrophy. The results of his physical examination were consistent with lingual tonsil hypertrophy as well as with an elongated soft palate. He under-
went a lingual tonsillectomy and soft palate reduction. The lingual tonsils were reduced with a coblation tech-
nique (ArthroCare ENT, Sunnyvale, California) at a set-
ning of 9 coblation and 5 coagulation. The soft palate was
partially excised and reduced with electrocautery at a set-

Abbreviations: Ad, adenoidectomy; LT, lingual tonsillectomy.
a One patient underwent LT alone and again as part of a multilevel,
single-stage procedure at a later date.

<table>
<thead>
<tr>
<th>Variable</th>
<th>OPS</th>
<th>Non-OPS</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Multilevel, single-stage procedure (49 patients)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Posterior midline glossectomy, coblation</td>
<td>1</td>
<td>7</td>
<td>8</td>
</tr>
<tr>
<td>Radiofrequency tongue ablation</td>
<td>1</td>
<td>4</td>
<td>5</td>
</tr>
<tr>
<td>LT, coblation</td>
<td>2</td>
<td>36</td>
<td>38</td>
</tr>
<tr>
<td>LT, electrocautery</td>
<td>2</td>
<td>8</td>
<td>10</td>
</tr>
<tr>
<td>LT, microdebrider and electrocautery</td>
<td>0</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>Ad, coblation</td>
<td>0</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>Ad, electrocautery</td>
<td>2</td>
<td>35</td>
<td>37</td>
</tr>
<tr>
<td>Tonsillectomy</td>
<td>0</td>
<td>13</td>
<td>13</td>
</tr>
<tr>
<td>Revision tonsillectomy, inferior palatine region</td>
<td>1</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>Uvulopalatopharyngoplasty</td>
<td>0</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Soft palate reduction</td>
<td>1</td>
<td>4</td>
<td>5</td>
</tr>
<tr>
<td>Total No. of procedures</td>
<td>121</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Table 1. Distribution of Procedures, Broken Down by Multilevel, Single-Stage Procedure vs Single-Level, Single-Stage Procedure and Presence or Absence of Oropharyngeal Stenosis (OPS)4

A one patient underwent LT alone and again as part of a multilevel,
single-stage procedure at a later date.
ting of 12-W spray. The technique used was a modification of the Z-palatoplasty described by Friedman et al.9 Modification included uvulectomy.

Patient 2 had 3 subsequent office visits, with no OPS noted. However, scarring of the palate and narrowing of the posterior oral airway, more on the right side than on the left, were noted at a fourth office visit 7 months after surgery. The patient underwent a right pharyngoplasty. Needlepoint electrocautery was used to divide a scar band, which was present between the right side of the soft palate, the right lateral pharyngeal wall, and the tongue base. The edges of this incision were closed to minimize exposed raw surfaces.

The patient was seen in clinic twice after surgery, with noted improvement of his OPS on the right side. However, 10 months later, he was found to have recurrent OPS with worsening of his OSA and a new oxygen requirement. Revision pharyngoplasty was performed as a horizontal-to-vertical pharyngoplasty (Figure) according to the technique described by McLaughlin et al.2 The goal of this technique is to lengthen the distance between the tongue base and the palate, to reorient the vector of scar tissue, and to minimize exposed raw tissue. A horizontal (posterior to anterior) incision was made in the scar tissue bilaterally to release it, exposing the underlying pharyngeal musculature. The muscle appeared to contribute to the OPS and was cut as well. Next, the released scar tissue was closed in a vertical fashion, lengthening the distance between the soft palate and the tongue base. Triamcinolone (40 mg) was injected into the lateral pharyngeal tissue and lateral tongue base region, mostly on the right side.

The following month, the patient underwent another examination under anesthesia along with triamcinolone injection into bilateral pharyngeal scar tissue. He was then seen in clinic 1 month later and noted to be significantly improved. He no longer required oxygen and was awaiting another sleep study.

In the early 20th century, OPS was commonly attributed to infection of the oropharynx. As cases of OPS attributable to infectious origins have declined because of antibiotic therapy, surgery has emerged as the most common cause. While OPS may result from T±A, it remains a rare complication, and many contemporary accounts of adenontonsillectomy complications do not mention it.7,9 Multiple risk factors, including excessive cautery, bleeding, a predisposition to keloid formation, and deep dissection in the region of the inferior tonsillar poles with removal of adjacent lingual tonsil tissue, have been proposed.1,2 Most reported cases have occurred in children.1,2

A review of the literature on uvulopalatopharyngoplasty, lingual tonsillectomy, and tongue base reduction procedures performed alone did not show evidence of OPS.1,10-18 Lin and Koltaï19 noted the potential for adhesions between the epiglottis and the tongue base after coblation lingual tonsillectomy and suggested additional caution during ablation of lingual tonsil tissue in the vallecula.

Similarly, many authors have reported no evidence of OPS after multilevel, single-stage upper airway procedures involving radiofrequency tongue base reduction.3,20-22 Although Ceylan et al30 speculate that OPS may occur after multilevel, single-stage procedures that address the palatine tonsils, base of tongue, and soft palate, their review of 47 patients treated with simultaneous radiofrequency tongue base and palate reduction noted no stenosis during 12 months of follow-up. Stuck et al31 reviewed 258 procedures that combined radiofrequency tongue base reduction and soft palate or oropharyngeal procedures. None of the patients involved developed OPS during a mean follow-up of approximately 3 months. These studies involved adult patients.

Table 2. Clinical Courses of 4 Patients With Oropharyngeal Stenosis (OPS)

<table>
<thead>
<tr>
<th>Patient No./Age at Multilevel, Single-Stage Procedure</th>
<th>Indication</th>
<th>Multilevel, Single-Stage Procedure</th>
<th>Duration Until OPS Noticed, mo</th>
<th>Surgical Repair</th>
<th>Triamcinolone, No. of Injections</th>
<th>Complication</th>
<th>Follow-up, mo</th>
</tr>
</thead>
<tbody>
<tr>
<td>1/5 y 5 mo UAO, LTH, Glossoptosis</td>
<td>Adenoectomy, electrocautery</td>
<td>3</td>
<td>Left pharyngoplasty</td>
<td>3</td>
<td>CPAP</td>
<td></td>
<td>24</td>
</tr>
<tr>
<td>2/3 y 5 mo OSA, LTH, Elongated soft palate</td>
<td>Soft palate reduction, electrocautery</td>
<td>7</td>
<td>Scar division and mucosal flaps</td>
<td>2</td>
<td>Continued OSA</td>
<td></td>
<td>21</td>
</tr>
<tr>
<td>3/3 y 4 mo Chronic lingual tonsillitis</td>
<td>Revision lingual tonsillectomy, lymphoid tissue reduction in palate tonsil region and posterior pharyngeal wall, electrocautery</td>
<td>10</td>
<td>Bilateral H-V plasty</td>
<td>2</td>
<td>Intraoperative scar division</td>
<td></td>
<td>11</td>
</tr>
<tr>
<td>4/5 y 8 mo UAO, LTH, Glossoptosis</td>
<td>Adenoectomy, electrocautery</td>
<td>4</td>
<td>Bilateral H-V plasty, partial uvulectomy, Right H-V plasty</td>
<td>3</td>
<td>Delayed decannulation</td>
<td></td>
<td>13</td>
</tr>
</tbody>
</table>

Abbreviations: CPAP, continuous positive airway pressure; H-V plasty, horizontal-to-vertical pharyngoplasty; LTH, lingual tonsil hypertrophy; OPS, oropharyngeal stenosis; OSA, obstructive sleep apnea; RFA, radiofrequency ablation; UAO, upper airway obstruction.
Many other authors have reported no OPS after multilevel, single-stage procedures involving techniques with more extensive mucosal incisions than radiofrequency tissue ablation.4,12,16,23-25 These studies were conducted in adults. The procedures often combined surgery of the tongue base or lingual tonsillectomy with soft palate surgery, adenoidectomy, or tonsillectomy and are therefore similar to the procedures that our series of patients underwent. We were able to find 1 patient with OPS who had undergone a multilevel, single-stage procedure other than adenotonsillectomy. Interestingly, this patient was an adult who underwent a lingual tonsillectomy combined with palatine tonsillectomy and required extensive tissue dissection and silver nitrate cauterization.1

Our patients with OPS were all young children, 5 1/2 years younger than our multilevel, single-stage series average. It is likely that a young age and correspondingly small upper airway elevate the risk of OPS. When used in these small spaces, surgical techniques can incite inflammation and injure the mucosa beyond the point at which the instrument is placed. When additional levels within a small upper aerodigestive tract are addressed, this risk is likely compounded.

It is worth noting that we have not seen OPS in our routine adenotonsillectomy population, a population that includes young children with smaller upper airways. It is our practice to use electrocautery settings as high as 36 W to perform adenoidectomy. We have not experienced nasopharyngeal stenosis or OPS in this population, despite the close proximity of raw or injured mucosa in the nasopharynx and the oropharynx. It is probable, therefore, that the distance between the adenoid bed or soft palate and the lingual tonsil region (the operative sites in patients 1, 2, and 4) would be sufficient to avoid this complication, even in small children. However, the presence of OPS in our series and the apparent rarity of this complication in the adult and older children populations raises the concern that the inherently smaller upper airways of young children and the resulting closer approximation of raw or injured mucosa is a risk factor for OPS when lingual tonsillectomy is performed as part of a multilevel, single-stage procedure.

Oropharyngeal stenosis was a delayed complication in our patients, presenting 2 to 8 months after surgery and after examination results were normal. Symptoms ranged from the mild or nonexistent to the severe. Previous reports of OPS have also noted the delayed nature and varied presentation of this complication.1,2 Because of the unpredictable nature of OPS, follow-up of patients undergoing multilevel, single-stage procedures that include lingual tonsillectomy is mandatory.
sillecctiony should also include examination of the oropharynx and tongue base, ideally with both an oral examination and nasopharyngoscopy to give a better view of the tongue base. The examination should be performed 3 to 6 months after the procedure. Caregivers should be warned of symptoms and signs that should prompt an office visit to an otolaryngologist.

Various repair techniques have been described.\textsuperscript{13} The most recently reported techniques use surgery and traimcinoi injections, alone or in combination.\textsuperscript{12,13} McLaughlin et al\textsuperscript{2} concluded that trimcinoi injection alone may suffice for OPS. They reserved horizontal-to-vertical pharyngoplasty for severe OPS that failed to respond to trimcinoi injection alone. We have chosen to follow that lesson, combining trimcinoi injection with pharyngoplasty of some form in all severe cases. Our case with more minor scar formation has been treated with scar division only. We are cautiously optimistic regarding our success to date and continue close follow-up of our patients.

In conclusion, surgery is now the most common cause of OPS, which was previously attributed to infection. Historically, tonsillecctiony and, more commonly, adenotonsillecctiony (a multilevel, single-stage procedure) have been associated with OPS. The frequency of this particular complication after T&A is exceedingly low. The risk appears to be similarly low in adults who undergo other types of multilevel, single-stage airway surgery. However, in young children who undergo multilevel, single-stage procedures that include lingual tonsillecctiony, OPS represents a significant potential complication, as our OPS rate of 8.2% demonstrates. Furthermore, OPS is a delayed, potentially asymptomatic complication. It may be successfully managed in the operating room. The risk of this complication should be discussed with parents or guardians. Although families frequently wish to minimize the number of separate surgical procedures for their children, staging of multilevel procedures that include lingual tonsillecctiony should be considered. For those patients who do undergo multilevel, single-stage procedures that include lingual tonsillecctiony, appropriate follow-up is suggested.

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Author Contributions: Dr Shott had full access to all the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis. Study concept and design: Prager. Acquisition of data: Prager. Analysis and interpretation of data: Prager, Hopkins, Propst, Shott, and Cotton. Drafting of the manuscript: Prager, Hopkins, Propst, and Shott. Critical revision of the manuscript for important intellectual content: Prager, Hopkins, Propst, Shott, and Cotton. Statistical analysis: Prager and Propst. Administrative, technical, and material support: Hopkins. Study supervision: Shott and Cotton.

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REFERENCES