



The role of laryngotracheal reconstruction in the management of recurrent croup in patients with subglottic stenosis



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ABSTRACT

Objectives: To determine the role of laryngotracheal reconstruction for recurrent croup and evaluate surgical outcomes in this cohort of patients.

Methods: Retrospective chart review at a tertiary care pediatric hospital.

Results: Six patients who underwent laryngotracheal reconstruction (LTR) for recurrent croup with underlying subglottic stenosis were identified through a search of our IRB-approved airway database. At the time of diagnostic bronchoscopy, all 6 patients had grade 2 subglottic stenosis. All patients were treated for reflux and underwent esophageal biopsies at the time of diagnostic bronchoscopy; 1 patient had eosinophilic esophagitis which was treated. All patients had a history of at least 3 episodes of croup in a 1 year period requiring multiple hospital admissions. Average age at the time of LTR was 39 months (range 13–69); 5 patients underwent anterior graft only and 1 patient underwent anterior and posterior grafts. Patients were intubated for an average of 5 (range 3–8) days and hospitalized for an average of 12 (range 7–20) days post-operatively. One patient experienced narcotic withdrawal post-operatively, but there were no other post-operative complications. All patients underwent follow-up airway endoscopy within 4 weeks and none required any further dilation procedures. Average post-operative follow-up was 24 months (range 10–48) and none of the patients experienced any further episodes of croup.

Conclusions: Single stage LTR is a safe and effective treatment for recurrent croup in the setting of underlying subglottic stenosis, and should be considered in patients who are refractory to medical management.

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1. Introduction

Viral croup is an infectious process involving the upper airway and represents the most common cause of upper airway obstruction in children aged 6 months to 6 years [1]. The parainfluenza virus accounts for the majority of cases, however other viruses including respiratory syncytial virus and influenza virus have also been implicated [2]. Typical presenting symptoms are hoarseness, barking cough and stridor; severe respiratory distress resulting in hospitalization is uncommon, occurring in less than 3% of cases [3].

Recurrent croup is significantly less common than acute viral croup, and has been estimated at a prevalence of approximately 6%

in children younger than age 4 [4]. Multiple recent studies have highlighted the importance of operative endoscopy in this cohort of patients to evaluate for underlying pathology [3,5,6]. All of these studies found high rates of reflux disease in these patients, ranging from 26 to 82% [3,5], as well as a significant percentage of patients with underlying subglottic stenosis, ranging from 18% to 100% [3,5,6]. These findings are consistent with a recent study published at our institution, which found that 20.8% of patients who presented with recurrent croup had underlying subglottic stenosis [7]. Other findings reported on operative endoscopy include eosinophilic esophagitis, tracheomalacia, bronchomalacia, laryngomalacia, and other less common upper airway abnormalities. Given the wide variety of possible underlying etiologies for this complex problem, a multidisciplinary approach to work-up and management of these patients has been recommended [8].

Management of recurrent croup typically involves maximally treating any underlying conditions medically. For patients who

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have been identified with subglottic stenosis, the initial medical management typically involved reflux medications due to the strong association of subglottic stenosis and reflux disease [9]. An algorithm suggested by Kwong et al. recommends starting reflux medications in all recurrent croup patients who have subglottic stenosis on bronchoscopy, with reassessment following 1 month of reflux medications. In their cohort of patients, 86% improved or resolved their recurrent croup episodes following initiation of reflux medications [5].

Further management for those patients with recurrent croup and subglottic stenosis who do not respond favorably to reflux medications alone is not as well-described in the literature. Although single stage laryngotracheal reconstruction has been well established for management of subglottic stenosis in general [10,11] its role in the management of recurrent croup in these patients is less clear. Herein, we share our experiences with airway reconstruction in this cohort of patients to help delineate the role of laryngotracheal reconstruction in the management of recurrent croup.

2. Methods

Six patients who underwent laryngotracheal reconstruction for recurrent croup from 2010 to 2014 were identified through a search of our IRB-approved airway database and included in this retrospective study. Inclusion criteria included patients who had been evaluated in our airway clinic for recurrent croup and had diagnostic bronchoscopy confirming underlying subglottic stenosis. All patients had undergone bronchoscopy with sizing of the airway to estimate the percentage of subglottic stenosis. This was done by sizing the airway using endotracheal tubes and calculating the grade of stenosis using the well-known Cotton-Myer grading scale [12]. Exclusion criteria included any patients who did not have pre-operative bronchoscopy findings available, or who did not undergo laryngotracheal reconstruction as well as patients with multilevel airway disease. One patient did have a history of tracheal vascular compression but had previously undergone aortopexy with no current significant vascular compression and she was therefore included. Charts were reviewed for comorbidities, severity/frequency of pre-operative croup episodes, surgical technique, degree of pre-operative subglottic stenosis, post-operative course and follow-up.

3. Results

Six patients who underwent laryngotracheal reconstruction for recurrent croup were identified. Average age at time of surgery was 39 months (range 13–69), and all patients had a grade 2 subglottic stenosis. All patients were treated with reflux medications prior to consideration for laryngotracheal reconstruction, and all patients had esophageal biopsies to rule out eosinophilic esophagitis. We do not routinely use any other anti-inflammatory medications or antibiotics in recurrent croup patients. No patients underwent immunological work-up. One patient was diagnosed with eosinophilic esophagitis; this prompted a gastroenterology referral and she was controlled with reflux medications and dietary changes. She had repeat esophageal biopsies which were negative for eosinophilia prior to her airway reconstruction. The remaining five patients were not evaluated by gastroenterology. All six patients had pre-operative modified barium swallow studies, and none exhibited aspiration. Four of the 6 patients were ex-premature infants with a history of intubation, one patient had a history of a tracheotomy in the past for respiratory failure following surgery for spina bifida, one had a history of tracheal vascular compression which had previously been addressed with aortopexy, and one patient was healthy with the exception of asthma. Patient

Table 1
Patient characteristics.

Patient	Age (months)	Comorbidities	Grade of SGS
1	60	Full-term, spina bifida, h/o trach	2
2	28	ex-32 weeker, intubated in NICU, h/o vascular compression s/p aortopexy	2
3	69	Full-term, asthma	2
4	31	ex-28 weeker, intubated in NICU	2
5	32	ex-30 weeker, intubated in NICU, BPD	2
6	13	ex-27 weeker, intubated in NICU	2

NICU = neonatal ICU.

characteristics are shown in Table 1. All patients had persistent croup episodes requiring frequent emergency room visits and hospital admissions. Frequency of croup episodes and quantity of emergency room visits and hospital admissions in the year prior to surgery are shown in Table 2. All documented croup episodes were treated with steroids.

All patients underwent single-stage laryngotracheal reconstruction; 3 had anterior thyroid ala grafts, 2 had anterior costal cartilage grafts; 1 had anterior and posterior costal cartilage grafts. Patients were intubated for an average of 5 days (range 3–8) and hospitalized for an average of 12 days (range 7–20). There were no major post-operative complications and one minor post-operative complication, which was narcotic withdrawal in one patient. Interval bronchoscopy was performed within 5 weeks in all patients; all airways were graded as normal and no adjunctive procedures were performed. Average duration of post-operative follow-up is 23 months (range 10–48) and none of the patients have had any additional episodes of croup. Other than peri-operative steroids, none of the patients required any courses of steroids for airway symptoms post-operatively. These findings are summarized in Table 3.

4. Discussion

Recurrent croup can be a challenging disease process to manage, particularly in patients who have underlying subglottic stenosis. Given the high incidence of concomitant reflux disease [3,5] which may contribute to subglottic narrowing in these patients, reflux management is paramount in these patients. It has also been our practice to biopsy the esophagus at the time of bronchoscopy to rule out eosinophilic esophagitis, which although rare can contribute to significant laryngeal and subglottic inflammation. One of the patients in this series had a positive biopsy for eosinophilic esophagitis; she was referred to gastroenterology, treated, and had repeat negative biopsies prior to being a candidate for airway surgery. If recurrent croup does not improve despite medical management of reflux disease and eosinophilic esophagitis, patients are potential candidates for airway surgery.

Five of our patient had risk factors for subglottic stenosis—4 were ex-premature infants who spent significant time in the

Table 2
Croup episodes in one year preceding LTR.

Patient	No. of croup episodes	No. of ED visits	No. of hospitalizations
1	4	3	2
2	7	2	1
3	9	5	1
4	5	4	0
5	9	3	1
6	4	3	1

ED = emergency department.

Table 3
Operative and post-operative course.

Patient	Procedure	Days intubated	Days hospitalized	Post-op complications	Interval to surveillance bronch (weeks)	Post-op croup episodes	Adjunctive procedures	Duration of follow-up (months)
1	A+P costal graft	7	14	None	4	None	None	48
2	A costal graft	8	20	Withdrawal	4	None	None	36
3	A costal graft	3	7	None	5	None	None	18
4	A thyroid ala graft	4	8	None	3	None	None	10
5	A thyroid ala graft	7	11	None	4	None	None	10
6	A thyroid ala graft	3	10	None	4	None	None	24

A = anterior; P = posterior.

neonatal intensive care unit and were intubated and one had a prior history of a tracheotomy but had been decannulated; the sixth patient had congenital subglottic stenosis. Neonatal intubation is a well-known risk factor for subglottic stenosis, although the incidence of subglottic stenosis in these children has decreased significantly since the 1990's, likely due to increased awareness and improved techniques of handling these neonates [13].

Although these patients have relatively mild subglottic stenosis, in the grade 2 range and are generally asymptomatic at baseline, they can become quite symptomatic with upper respiratory tract infections. This requires frequent trips to the emergency room and can be very stressful and scary for families, particularly those who do not live in close proximity to a children's hospital. One of our patients lives in a remote part of Pennsylvania and had to be "life-flighted" to our hospital on multiple occasions for treatment of croup. All of the patients in our series had at least 4 episodes of croup during the year prior to airway reconstruction, as well as multiple emergency room visits and/or hospitalizations. Although there are no studies to date looking at parental quality of life in recurrent croup patients, it has been our experience that many families experience significant distress as a result of their child's recurrent croup. Single-stage LTR provides a safe and effective treatment option for these patients.

Single-stage LTR is a well-described and accepted technique for management of subglottic stenosis [10,11], however it is not without complications. Its success rate is fairly high, generally reported at greater than 90% [10,11]. All of our patients had complete resolution of their recurrent croup, and there was only one post-operative complication in a child who had narcotic withdrawal. Major complications are uncommon, however these patients are at risk of pulmonary complications, prolonged hospitalization, and narcotic withdrawal due to sedation during the time of intubation [14]. Our practice has been to try to extubate by post-operative day 7, however patients can require prolonged intubation for various reasons.

In addition, post-operative follow-up is important in these patients, as there is a risk of granulation tissue formation or restenosis which may require adjunctive procedures. Although none of our patients required any adjunctive procedures, they all had a surveillance bronchoscopy approximately one month post-operatively to assess the status of the airway; frequency of need for adjunctive procedures for single-stage LTR is reported up to 60% in the literature [15], so it is imperative to perform surveillance bronchoscopy. Families and surgeons must weigh the quality of life issues related to the recurrent croup episodes as well as the frequency/severity of episodes with the potential complications of LTR in making the decision to proceed with surgery in these patients. Based on our experience, we recommend considering offering laryngotracheal reconstruction to children who have had at least 4 episodes of croup in a one-year period with significant quality of life concerns by the parents.

5. Conclusions

Single-stage LTR is a safe and effective management strategy for patients with recurrent croup with underlying subglottic stenosis who continue to have episodes of croup despite management of medical comorbidities. Recurrent croup can be very distressing to families, and single-stage LTR can be considered as a surgical option in certain cases.

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Conflict of interest statement

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