Original Research

AMERICAN ACADEMY OF OTOLARYNGOLOGY–HEAD AND NECK SURGERY FOUNDATION

Otolaryngology–
Head and Neck Surgery
1–6

DOI: 10.1177/0194599814567106
http://otojournal.org
SAGE

Comparison of Hybrid Laryngotracheal Reconstruction to Traditional Single- and Double-Stage Laryngotracheal Reconstruction

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No sponsorships or competing interests have been disclosed for this article.

Abstract

Objectives. (1) To describe outcomes from and modifications to the hybrid laryngotracheal reconstruction (LTR) technique and (2) to compare this technique to traditional single- and double-stage LTR (ssLTR/dsLTR).

Study Design. Chart review with case series.

Setting. Tertiary care otolaryngology specialty hospital.

Subjects. All patients under 18 years of age who underwent LTR by a single surgeon from July 1, 2009, to December 31, 2013.

Methods. Charts were assessed for age, gender, etiology of stenosis, type of reconstruction, comorbidities, length of stay, complications, and tracheostomy status. Analysis was performed using Kruskal-Wallis and Wilcoxon rank sum analysis.

Results. Forty-four patients were identified, with 13 hybrid LTRs, 27 ssLTRs, and 4 dsLTRs. Of the hybrid LTRs, an overall decannulation rate of 76.9% was noted, comparable to those for dsLTR. The hybrid LTR technique offered a significantly shorter period of narcotic use when compared to ssLTR (median 15 vs 21 days, $P < .01$). No patients in the hybrid LTR group developed supraglottic granulation tissue. There was no statistically significant difference in median length of stay for ssLTRs, dsLTRs, and hybrid LTRs ($P = .38$).

Conclusion. The hybrid LTR technique is well tolerated and useful in patients of all ages. Narcotics can be weaned more quickly due to the presence of a secure airway at all times via the existing tracheostomy. Use of a long stent prevents formation of granulation tissue that may be seen with a suprastomal stent. This technique should be considered in patients with high-grade stenosis with a preexisting tracheostomy.

Keywords

laryngotracheal reconstruction, LTR, subglottic stenosis, hybrid LTR

Received August 29, 2014; revised October 31, 2014; accepted December 15, 2014.

Introduction

Pediatric laryngotracheal stenosis is a problem frequently encountered by the pediatric otolaryngologist. The primary etiology has changed over time, from diphtheria in the early twentieth century, to posttraumatic injuries from motor vehicle accidents in the 1930s, to prolonged intubation in neonates beginning in the 1960s,[1] which led to an increase in incidence of subglottic stenosis.[2] Although the occurrence has decreased due to the improved airway management seen in the following 2 decades,[2] the incidence today is still approximately 0.63% to 2% when all etiologies, including postintubation and congenital stenosis, are taken into account.[3,4] During the neonatal period, anterior cricoid split and tracheostomy are certainly acceptable methods for management of subglottic stenosis.[3] For patients presenting after the neonatal stage, numerous options exist for treatment, including tracheostomy, endoscopic airway surgery, and open airway reconstruction. Despite advancement in endoscopic management, open techniques remain the mainstay for treatment, whether performed as a primary operation or following failure of endoscopic techniques.[2,5]

Two primary techniques of open laryngotracheal reconstruction (LTR) with cartilage grafting have been described. Double-stage LTR (dsLTR) involves keeping the tracheostomy tube in place following reconstruction and placement of a suprastomal stent in order to maintain a patent airway lumen while healing occurs. Planned decannulation then occurs at a later time. Advantages of this technique include ability to avoid prolonged postoperative sedation and presence of a stable airway in the form of a tracheostomy tube.

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The primary disadvantage is that complications related directly to the stent may occur, including suprastomal granulation tissue and stent migration. In addition, inappropriate stent length may result in aspiration (too long) or inadequate stenting of the airway (too short).[6]

In contrast, the ssLTR involves reconstructing the airway while decannulating the patient at the time of surgery (if a tracheostomy is present) or avoiding a tracheostomy altogether. Advantages of this technique include removal of tracheostomy tube at the time of surgery and use of the endotracheal tube as a single long stent. Drawbacks to this technique are the possible need for reintubation and the effects of prolonged sedation, including possible withdrawal and need for extensive physical therapy due to weakness.[7,8]

In 2013, Setlur et al[9] described a novel technique for LTR, called the hybrid, or one-and-a-half-stage, LTR. This method of reconstruction combines aspects of both the ssLTR and dsLTR, allowing for presence of a single long stent via the same endotracheal tube used for ventilation, as with the ssLTR, as well as a small stent in the tracheostoma fashioned from a cut endotracheal tube.[9] In our experience, the noted advantages have included lack of granulation tissue formation and presence of a safety valve should a patient need prolonged ventilation (ie due to withdrawal). Initially developed for use in airway surgical missions abroad, this technique was felt to bridge the gap between the riskiness of an ssLTR, with regards to postextubation safety in a less familiar medical environment, with the safety of the persistent tracheostomy of the dsLTR, but without a stent left in place to be removed after the original surgical team was out of the country. This was done with the plan of decannulation upon return the following year for another mission. The technique was received extremely favorably by the pediatric intensive care unit (PICU) team for its safety as well as by families who were anxious about respiratory difficulty in the case of withdrawal. Therefore, the technique was brought back to our home institution, and over the past 3 years, it has become more commonly utilized. We present outcomes from patients who have undergone hybrid LTR and compare this technique to traditional ssLTR and dsLTR.

Patients and Methods

Following approval for this study from the Institutional Review Board at the Massachusetts Eye and Ear Infirmary (MEEI), charts of all patients who had undergone laryngotracheal reconstruction by a single surgeon at MEEI between July 1, 2009, and December 31, 2013, were identified and reviewed. The medical record of each patient was reviewed for details in management and outcome. Specifically, data extracted included date of birth, sex, etiology and degree of stenosis, age at airway reconstruction, length of stay, comorbidities, surgical technique used, postoperative complications, tracheostomy status, and total length of narcotic use (including both inpatient use and outpatient prescriptions). Intravenous fentanyl and midazolam were used in the immediate postoperative period, with transition to morphine and lorazepam, given either by mouth (if the patient had passed a swallow study) or by nasogastic/gastrostomy tube. There was not a consistently accurate obtainment of pain scale levels for patients driving narcotic use; narcotics were primarily given for sedation, prevention of withdrawal, and treatment of withdrawal. Over the past 2 years, these medications have been administered based on a specific sedation protocol developed at our institution.[10] Narcotic use was not available for the all patients in the dsLTR group; therefore no significant comparison could be made with this group. Kruskal-Wallis and Wilcoxon rank sum analysis were used to compare outcomes between hybrid, single-stage, and double-stage LTRs.

Results

Patient Characteristics

Forty-four patients were identified who underwent laryngotracheal reconstruction between July 1, 2009, and December 31, 2013 (**Table 1**). Twenty-seven underwent ssLTR, 4 underwent dsLTR, and 13 underwent hybrid LTR. In the ssLTR and hybrid LTR groups, all patients underwent bronchoscopy at 1 week and 2 weeks postoperatively, followed by repeat bronchoscopy at 6 weeks postoperatively. In the dsLTR group, stent removal occurred at 1 week postoperatively, with repeat bronchoscopy 1 month postoperatively.

Of those who underwent ssLTR, 63% (17/27) had grade 2 stenosis, 33% (9/27) had grade 3 stenosis, and 3.7% (1/27) had grade 4 stenosis. The median age at the time of reconstruction was 23 months (interquartile range [IQR] = 16, 37]. In addition, 55.6% (15/27) were premature, 18.5% (5/27) had a history of chronic lung disease (CLD), and 18.5% (5/27) had gastroesophageal reflux (GERD). Seventy percent of these patients had tracheostomies prior to surgery. The median length of follow-up in these patients was 12 months (IQR = 2, 26.5).

Of the patients who underwent dsLTR, 75% (3/4) had grade 3 stenosis, and 25% (1/4) had grade 4 stenosis. The median age at surgery was 40.5 months (IQR = 13, 67.5). Two patients (50%) had a history of CLD. All patients had preexisting tracheostomies. The median length of follow-up in these patients was 42 months (IQR = 30.25, 48.5).

Of the patients who underwent hybrid LTR, 15.4% (2/13) had grade 2 stenosis, 61.5% (8/13) had grade 3 stenosis, and 15.4% (2/13) had grade 4 stenosis. One patient had bilateral true vocal fold paralysis. Median age at the time of reconstruction was 33 months (IQR = 20, 66). In addition, 53.8% (7/13) of patients had a history of prematurity, 30.8% (4/13) had a history of CLD, and 23.1% (3/13) had GERD. One hundred percent of these patients had tracheostomies preoperatively. The median length of follow-up in these patients was 22 months (IQR = 15, 27).

Postoperative Course

When comparing length of stay (**Table 2**), patients who underwent ssLTR, dsLTR, and hybrid LTRs had median lengths of stay of 15.5 days, 9.5 days, and 15 days, respectively (P = .38, Kruskal-Wallis test). However, in

Table 1. Patient Characteristics.

	Single-Stage LTR	Double-Stage LTR	Hybrid LTR
No. of patients	27	4	13
Males (%)	13 (48.1)	3 (75)	10 (76.9)
Median age at surgery in months (IQR)	23 (16, 37)	40.5 (13, 67.5)	33 (20, 66)
Grade of stenosis			
Grade 2 (%)	17 (63)	0 (0)	2 (15.4)
Grade 3 (%)	9 (33)	3 (75)	8 (61.5)
Grade 4 (%)	1 (3.7)	1 (25)	2 (15.4)
Pre-LTR tracheostomy (%)	19 (70)	4 (100)	13 (100)
Prematurity (%)	15 (55.6)	2 (50)	7 (53.8)
Chronic lung disease (%)	5 (18.5)	2 (50)	4 (30.8)
GERD (%)	5 (18.5)	0 (0)	3 (23.1)

Abbreviations: GERD, gastroesophageal reflux; IQR, interquartile range; LTR, laryngotracheal reconstruction.

Table 2. Outcomes Following LTR.

	Single-Stage LTR	Double-Stage LTR	Hybrid LTR	*P* Value
Median length of stay in days (IQR)	15.5 (14, 19)	9.5 (8, 17)	15 (14,18)	.38
Median length of narcotic use in days (IQR)	21 (17, 28)	—	15 (13, 19)	.007
Decannulated following surgery (%)	23 (85)	0 (0)	10 (76.9)	—
Median length of follow-up in months (IQR)	12 (2, 26.5)	42 (30.25, 48.5)	15 (22, 27)	

Abbreviations: IQR, interquartile range; LTR, laryngotracheal reconstruction.

comparing total length of narcotic use between ssLTR and hybrid LTR (including sedation and wean from sedation), a statistically significant difference was seen in the median values of 21 days (IQR = 17, 28) and 15 days (IQR = 13, 19), respectively ($P = .007$, Wilcoxon rank sum test). These data were not available for the dsLTR group because it was not consistently recorded.

Complications

In the ssLTR group, 4 patients (14.8%) required reintubation and subsequent tracheostomy placement due to prolonged ventilator dependence. The hybrid LTR group included 3 patients (23.1%) who required prolonged ventilator dependence: 2 for pneumonia and 1 for withdrawal. These patients were able to use their tracheostomy tubes, which had remained in place after removal of the endotracheal tube, for mechanical ventilation until they were able to support their own breathing. In the dsLTR group, 1 patient did develop granulation tissue above the stent in the glottic region. This patient ultimately required 2 additional surgeries prior to capping trials.

Additional Surgical Interventions

In the ssLTR group, 11 of 27 (40.7%) of patients underwent a total of 25 additional procedures following LTR, including balloon dilation, revision LTR, laser supraglottoplasty, and excision of subglottic granuloma in 1 patient. In the dsLTR group, 2 of 4 (50%) patients underwent a total of 10 additional procedures, including balloon dilation, laser supraglottoplasty, and repeat laryngofissure with stent replacement. In the hybrid LTR group, 7 of 13 (53.8%) patients underwent a total of 13 additional procedures, including balloon dilation, revision LTR, and endoscopic and open scar excision with placement of T-tube.

Decannulation

In the ssLTR group, operation-specific and overall decannulation/extubation rates were 85% and 92.6%, respectively. In the dsLTR group, 3 patients are currently undergoing capping trials, but none have been decannulated thus far. In the hybrid LTR group, the operation-specific and overall decannulation rate were 69.2% and 76.2%, respectively. At the time of this review, 1 additional patient was a candidate from an airway patency standpoint for decannulation but was unable to be decannulated quickly following the procedure due to CLD. Of those who could not be decannulated due to airway narrowing, 1 patient had severe tracheomalacia following a previous tracheoesophageal fistula repair, and another had severe structural damage to his airway following a motor vehicle accident, which led to restenosis following LTR. Despite repeat scar excision, this final patient had experienced extensive traumatic laryngeal injury and repeatedly restenosed following all additional interventions.

Discussion

The hybrid LTR technique was first described in 2013 by Setlur et al.[11] Patients who were felt to be the best candidates for this procedure were similar to those for dsLTR, namely, children predicted to require tracheostomy following surgery due to high grade stenosis, multilevel stenosis, and other comorbidities preventing decannulation.[11] In addition, parental apprehension with removal of tracheostomy tube may also be assuaged through use of this technique.[9]

One of the primary benefits of the hybrid LTR over the ssLTR is the presence of a safety valve in the form of the tracheostomy tube in the case of need for prolonged mechanical ventilation. This could be due to poor lung function or withdrawal from sedation. In our study, 3 patients who underwent hybrid LTR required repeat mechanical ventilation following extubation due to postoperative complications of pneumonia and withdrawal, which was easily achieved through use of the already present tracheostomy without need for reintubation. By having the tracheostomy tube in place, the PICU team could be more aggressive in their attempts to wean sedation safely, and the possible psychological distress for the parent that occurs from emergent reintubation was avoided. Because sedation could be weaned more quickly, the total length of narcotic use was significantly lower when we compared our patients who underwent ssLTR to those who underwent hybrid LTR (median 21 days vs 15 days, respectively; $P = .007$). There was no difference in length of stay in the hospital, but fewer patients were discharged with a prescription for a narcotic wean to be completed at home. Of note, though dsLTR patients typically have an average hospital stay of 3 to 5 days, our 4 patients had various reasons for the slightly extended stay, including feeding intolerance and parental discomfort with discharge with stent in place. Because there were only 4 patients in this group, the median value and lack of statistical significance are not generalizable. Those for the ssLTR and hybrid LTR appear to be more generalizable due to a greater number of patients.

When considering which patients are candidates for decannulation, many factors come into play, even if the airway is patent, including lung function, neurologic status, and parent comfort. Because the hybrid LTR is useful in the same subset of patients who are good candidates for dsLTR, decannulation rates could be expected to be in the same range as for that procedure, if equally efficacious. Our operation-specific decannulation rate was 69.2%, and the overall decannulation rate was 76.2%. In the first report comparing outcomes in ssLTR versus dsLTR, Saunders et al[12] demonstrated an operation-specific decannulation rate of 61.2% in the patients who underwent dsLTR. Hartnick et al[13,14] divided those who underwent dsLTR into groups based on Cotton-Myer grade, and their data showed that operation-specific decannulation rates for grade 2, 3, and 4 stenosis of 85%, 37%, and 50%, respectively. Most recently, in 2010, Smith et al[7] reported their outcomes from ssLTR and dsLTR, and the operation-specific decannulation rate demonstrated in their population was 68%. Therefore, our results thus far for decannulation are comparable to those in the literature. Further follow-up of these patients will allow for future comparison of overall decannulation rates.

The benefit of the hybrid LTR over dsLTR, however, is the decreased likelihood of granulation tissue formation due to presence of a long stent. By using the endotracheal tube to stent open the reconstruction instead of a suprastomal stent, placement of the cut edge of an endotracheal tube against a mucosal surface is avoided. In a recent study by Preciado[15] comparing silastic stents to Albouker stents in dsLTR, he reported the development of granulation tissue, albeit to varying extents, in all patients in the study. None of our patients who underwent hybrid LTR required revision surgery to remove granulation tissue.

When deciding which technique to use at our institution, ssLTR is still used consistently for children who present with lower grade stenosis, who do not have tracheostomies preoperatively, and who do not have comorbidities that would lead the surgeon to predict the need for a tracheostomy postoperatively. If a patient has a preoperative tracheostomy tube with a low-grade stenosis that could be addressed with either an ssLTR or a technique with delayed decannulation, weight is given to the parents' comfort level with decannulation at the time of surgery. In the case where the parent is not comfortable, the hybrid LTR has become the option of choice and has thus far been welcomed by the family. If given the choice of the dsLTR or the hybrid LTR, the trend has been to perform the hybrid LTR. However, the dsLTR still has utility in cases where a child may be too young or too sick to tolerate a significant amount of sedation/paralysis for a prolonged period of time, leaving him or her very weak and in need of extensive physical therapy, or when it is anticipated prolonged airway stenting may be indicated.

The primary weakness cited in Setlur et al's[9] report is the possible peristomal air leak while the patient is on the ventilator in the immediate postoperative period, creating possible problems in ventilating the patient. The author emphasizes the importance of a secure neck dressing to minimize the air leak. In order to improve on this limitation, a minor modification has been made. Instead of fashioning a tracheostomal stent from a cut endotracheal tube, it is now the practice of the primary surgeon to use a small tracheostomy tube, typically a 3.0 neonatal Shiley tracheostomy tube, to serve as the stent. The benefit of this change is 2-fold: the flanges of the tracheostomy tube allow the tube to rest flush with the skin of the neck and allow it to be easily secured with ties, as in a normal tracheostomy, thereby minimizing the surrounding leak. In addition, the tracheostomy tube and the endotracheal tube appear to occupy the airway simultaneously without difficulty, as the postreconstruction endoscopy has not shown any evidence of trauma, and it has shown excellent stenting of the reconstruction with the endotracheal tube. Second, in the rare but possible event of accidental extubation in the immediate postoperative period, the tracheostomy tube is already in place, allowing for ventilation to be continued until the proper otolaryngology personnel arrive to address the situation appropriately.

It is imperative that all members of the pediatric intensive care unit team understand the configuration of the endotracheal tube, the tracheostomy tube, and their relationships to the carina and to the site of reconstruction. Although both tubes share space in the trachea, we have not noted difficulty in accommodation of the 2. In addition, just as in the dsLTR, wound care of the tracheostomy tube site is imperative to prevent dehiscence of the cervical wound due to exposure of the incision to copious amounts of saliva/secretions.

Although we do feel that this new technique combines the beneficial aspects of the ssLTR and dsLTR (single long stent with decreased granulation tissue, presence of tracheostomy in case of airway emergency), the downside of combining these 2 techniques should be addressed as well. The hybrid LTR does use sedation and mechanical ventilation for the first week postoperatively, as done with the ssLTR. However, we feel that we have been able to keep narcotics to a minimum, particularly with the institution of our sedation protocol.[10] We did have 1 older child (15 years) who underwent hybrid LTR, and in this scenario, the patient was only given morphine as needed for pain control. Furthermore, we feel that the trade-off between lack of granulation tissue formation and need for sedation has been suitable. In addition, we do not feel that delayed decannulation is a drawback, as many of our patients need time to transition from tracheostomy dependence to respiratory independence due to other comorbidities, such as lung disease or hypotonia.

A unique noteworthy point about the hybrid LTR technique is the fact that it is the only technique of the 3 with a backup airway. In the ssLTR, if an endotracheal tube was to be inadvertently dislodged or the balloon were to rupture, an emergent, and possibly catastrophic, airway situation would ensue. With the dsLTR, if the stent were to migrate inferiorly due to excessive activity by the patient, the airway could become obstructed, and again, an airway emergency would result. When the hybrid technique is used, should something happen to the primary airway (endotracheal tube), the tracheostomy tube is immediately present and can help prevent an emergent situation.

While the ssLTR is still the technique of choice at our institution in patients with particular characteristics such as lack of comorbidities, lack of tracheostomy preoperatively, and lower grade stenosis, in patients who would traditionally be dsLTR candidates, the hybrid LTR has largely replaced the dsLTR primarily due to its safety, overwhelming parental and intensivist support, and lack of granulation tissue formation. This technique is a useful addition to the armamentarium of open airway reconstruction procedures.

While our data are promising, we recognize limitations in our study. First of all, the number of patients who underwent hybrid LTR is small, and the follow-up period is short. A larger study with longer follow-up is needed to assess the durability of this technique. In addition, the generalizability of the technique depends heavily on the comfort and ability of the intensivists caring for the patients postoperatively. At our institution, a strong relationship with the PICU team has been established over several years, allowing us to try this new technique safely.

Conclusion

The hybrid LTR appears to be a safe and successful technique for open airway reconstruction in carefully selected patients. It is particularly useful in those patients who are predicted to be tracheostomy-dependent following the procedure. Benefits of this technique include the presence of a long stent, avoiding formation of granulation tissue, and presence of the tracheostomy tube for ventilation if needed in an emergency situation.

Author Contributions

Nikhila Raol, conception/design, acquisition/analysis of data, interpretation of data, drafting of work, critical revision, final approval; **Derek Rogers**, conception/design, critical revision, final approval; **Jennifer Setlur**, conception/design, critical revision, final approval; **Christopher J. Hartnick**, conception/design, critical revision, final approval.

Disclosures

Competing interests: None.

Sponsorships: None.

Funding source: None.

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