**Growth and Management of Repaired Complete Tracheal Rings after Slide Tracheoplasty**

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**Abstract**

**Objective.** This study documents the growth and course of repaired complete tracheal rings over time after slide tracheoplasty.

**Study Design.** Case series with review.

**Setting.** Tertiary pediatric academic medical center.

**Subjects/Methods.** Medical records of pediatric patients with confirmed tracheal rings on bronchoscopy who underwent slide tracheoplasty between January 2001 and December 2015 were reviewed. Patients who had operative notes documenting tracheal sizing over time were included. Exclusion criteria included tracheal stenosis not caused by complete tracheal rings, surgical repair prior to presentation at our institution, or lack of adequate sizing information. The postoperative follow-up was examined and airway growth over time documented.

**Results.** Of 197 slide tracheoplasties performed during the study time period, 139 were for complete tracheal rings, and 40 of those children met inclusion criteria. The median age at time of surgery was 7 months, and the median initial airway size was 3.9 mm \((n = 34)\). The median growth postoperatively was 1.9 mm over a median follow-up period of 57 months \((0.42 \text{ mm/year})\), which is similar to growth rates of unrepaired complete tracheal rings \((P = .53)\). Children underwent a median of 10 postoperative endoscopies, with time between endoscopies increasing further out from surgery. The most commonly performed adjunctive procedure was balloon dilation.

**Conclusions.** This is the first study documenting continued growth of repaired complete tracheal rings after slide tracheoplasty. Postoperative endoscopic surveillance ensures adequate growth. Intervals between airway endoscopies can be increased as the child gets older, as the airway increases in size, and as long as symptoms are minimal.

**Keywords**

pediatric airway, complete tracheal rings, airway growth, management, airway sizing

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Tracheal stenosis in the pediatric population can be divided into congenital and acquired causes. Congenital tracheal stenosis is estimated to occur in only 1 in 64,500 cases\(^1\) and accounts for only 0.1% to 0.3% of all laryngotracheal stenosis cases.\(^2\) Despite being the most common form of congenital tracheal stenosis, complete tracheal rings are still extremely rare and account for \(<1\%\) of all airway stenosis.\(^2\) Complete tracheal rings result from a defect in embryogenesis after the eighth week of gestation, causing a complete cartilaginous ring with absence of the usual posterior membranous portion of the trachea. This may result in tracheal narrowing and subsequent symptomatology.

Most children with complete tracheal rings present in infancy with biphasic “washing-machine” stridor, retractions, increased work of breathing, and cyanotic spells and require early surgical repair.\(^3\) There are often comorbid conditions.
conditions such as a pulmonary artery sling, tracheal bronchus, Down syndrome, and variations of the VATER/VACTERL (vertebral, anal, cardiac, tracheoesophageal fistula, rectal, limb) spectrum. While it is the exception, a subset of patients presents either incidentally (eg, when being intubated for another reason) or with mild symptoms (eg, dyspnea on exertion). Some may even present as young adults with asthma-like symptoms. While previous reports have shown approximately 17% of patients with complete tracheal rings can be managed conservatively, most children will require surgical repair.4

The method of surgical repair for complete tracheal rings has evolved over the past several decades. Primary resection with reanastomosis,5 augmentation tracheoplasty (with costal cartilage,6-8 pericardial patch,9-11 or tracheal autograft12), tracheal allografting,13-15 balloon dilation,16 and laser division have all been described and successfully employed. However, since it was first described by Tsang and popularized by Grillo, slide tracheoplasty has emerged as the largely undisputed method of choice for repairing complete tracheal rings as it avoids the problems of excessive tracheal shortening with anastomotic tension, circumferential scarring, loss of mucociliary clearance, graft prolapse, and granulation around grafts.18-20 Grillo et al previously reported adequate growth after repair in several of these patients, but actual documentation of the rate of growth over time has not been described.19,21 This study documents the growth of repaired complete tracheal rings (RCTRs) longitudinally and details the postoperative course after slide tracheoplasty.

Methods

A case series with chart review was conducted on all patients presenting from January 1, 2001, to December 31, 2015, with congenital tracheal stenosis caused by complete tracheal rings. Patients ages 0 to 18 years with confirmed tracheal rings on microlaryngoscopy and bronchoscopy (MLB) who underwent slide tracheoplasty for repair of their complete tracheal rings were included if they had operative notes documenting formal tracheal sizing on at least 2 separate occasions spanning a 2-year postoperative course. Patients were identified from 2 sources. A previously created database documenting repair of complete tracheal rings from 2001 to 2014 was used to identify the majority of the patients. For the remaining year, a search was performed through the electronic medical record (EPIC; Epic Systems Corporation, Verona, Wisconsin) to capture all children who underwent slide tracheoplasty. Review of the medical record for each child found from these sources was employed to confirm the diagnosis of complete tracheal rings and the surgical repair with slide tracheoplasty. Exclusion criteria included tracheal stenosis not caused by complete tracheal rings, presentation at >18 years of age, surgical intervention for complete tracheal rings prior to presenting to our institution, or those who did not have adequate tracheal sizing documentation. Demographic information, comorbidities, other airway anomalies, symptoms, airway interventions, and information regarding the complete tracheal rings at each MLB were recorded. Long-segment vs short-segment complete tracheal rings were defined as encompassing >50% or <50% of the length of the trachea, respectively. Institutional review board approval was granted by Cincinnati Children’s Hospital Medical Center and the University of Cincinnati College of Medicine.

Sizing of the airway at the level of the complete tracheal rings, both before and after slide tracheoplasty, was performed using rigid bronchoscopy and uncuffed endotracheal tubes. The endotracheal tubes were placed under endoscopic visualization to ensure the narrowest portion of the trachea was sized. The endotracheal tube was loaded over a small Hopkins rod endoscope, with the tip of the tube being placed through the narrowest portion of the airway. The telescope was removed from the endotracheal tube and then reinserted outside the endotracheal tube to perform a leak test under endoscopic visualization. Airway size was determined by taking the outer diameter (in millimeters) of the largest endotracheal tube with a leak at less than 20 cm of water (Table 1). If a 2.0 endotracheal tube was too large for the airway but a smaller telescope could be passed, then the diameter of the telescope was used to estimate the airway size. In some instances, the small endotracheal tubes were not long enough to reach the distal trachea. In these cases, a modified endotracheal tube was created from 2 uncuffed endotracheal tubes as previously described elsewhere.4 The airway growth over time was defined by the change in airway diameter throughout postoperative follow-up based on this sizing method. Of note, the airway may not have been sized at every MLB if there was concern for acute exacerbation of symptoms, interfering with healing, or the airway growth was obviously adequate.

Descriptive statistics were calculated on demographics and clinical characteristics, including frequencies and percentages for categorical variables and medians with interquartile ranges (IQRs) for continuous variables. Histograms and boxplots were used to evaluate distributions for normality, and t tests were used to compare means of change in airway size before and after surgery as well as growth rates per year in repaired and unrepaired complete tracheal rings. Correlations between MLB interval (months) and airway size or age were tested using Spearman’s ρ correlation. Airway growth per year followed was calculated using the first measure after surgery and last measure of airway size divided by the overall time between those measurements per individual. Thus, the calculation of airway growth over time excluded the expansion of the airway due to the surgery itself. A P value of <.05 was considered significant. SAS version 9.4 (SAS Institute, Cary, North Carolina) was used for all analyses.

Results

In the study time period, 197 patients underwent slide tracheoplasty, with 139 of those having complete tracheal rings. Other indications for slide tracheoplasty are listed in
The airway diameter increased a mean of 1.1 mm after surgery and had a median initial airway size of 3.9 mm. Concurrent cardiovascular surgery procedures included repair of atrial septal defect (n = 4), ventricular septal defect repair (n = 4), atrial septal defect repair (n = 5), and pulmonary artery sling repair (n = 12), patent ductus arteriosus (n = 3) and tetralogy of Fallot (n = 3) being the most common, and 5 patients had prior cardiovascular surgery for congenital defects, with pulmonary artery sling (n = 3) and tetralogy of Fallot (n = 3) being the most common findings. Twenty-two patients underwent repair of congenital stenosis, with pulmonary artery sling (n = 12) and absent tracheal cartilage (n = 7) being the most frequent concurrent cardiovascular surgery procedures.

Patients underwent slide tracheoplasty at a median age of 7 months and had a median initial airway size of 3.9 mm. The airway diameter increased a mean of 1.1 mm after surgery or 56% (Figure 2). The pre- and postslide tracheoplasty airway sizes were significantly different by paired t test (P = .007). Preoperative sizing information was not available for 6 of the patients. The median airway growth after surgery was 1.9 mm over a median follow-up period of 57 months (0.42 mm/year) (P = .53). Increasing airway size was strongly positively correlated with increasing age (r = 0.75, P < .0001, Figure 3). The individual growth patterns over time are depicted in Figure 4.

Children underwent a median of 10 postoperative endoscopies, with time between endoscopies with sizing increasing as children got further out from surgery (r = 0.75, P < .001). The timing of postoperative airway endoscopy, both with and without sizing, is further detailed in Table 3. The most common adjunctive procedure was balloon dilation, which was performed a total of 23 times in the subject group. Nine children underwent a median of 2 balloon dilations each, with the first typically occurring within 1 month of surgery. Further adjunctive postoperative airway procedures are documented in Table 4.

Of the children who did not show significant growth over the long-term follow-up, several (n = 3) were asymptomatic and were followed serially without interventions. One patient underwent Palmaz stent placement in the early postoperative period. This was removed after 1 month, and he remained asymptomatic despite minimal change in airway diameter. Four patients were symptomatic with the following findings and underwent additional intervention: proximal anastomotic stenosis requiring revision slide tracheoplasty (n = 1), dyspnea and air trapping in a tracheal bronchus requiring placement of a Palmaz stent and demucosalization and cautery of the tracheal bronchus (n = 1), and suprastomal collapse (n = 1) and subglottic stenosis (n = 1) requiring laryngotracheoplasty.

### Discussion

This study demonstrates that growth of RCTRs generally does occur over time; however, the pattern may be variable and exceptions may occur. While growth occurs at approximately 0.42 mm, or about two-thirds an endotracheal tube size, per year, it is interesting to note that some children had several MLBs showing no growth or even decreased size from prior evaluations. In the early postoperative period, swelling and granulation tissue may have contributed to this. Throughout follow-up, a true lack of growth, edema from airway manipulation prior to sizing (eg, flexible bronchoscopy), variability in sizing technique, proximal airway abnormalities, or figure-of-8 deformities may have contributed to the findings.

Airway growth has previously been documented in unrepaired complete tracheal rings and reported in congenital tracheal stenosis repaired with either slide tracheoplasty or tracheal resection and anastomosis; however, this is the first and largest study to characterize this growth over time in patients with RCTRs. Our study’s mean growth rate of 0.42 mm per year is similar to the expected change in endotracheal tube size with age in children with normal tracheas based on standard endotracheal tube sizing formulas.
Also interesting is that while the airway size increased a mean of 56% when sized within a 3-month period after surgery, which is consistent with Grillo’s findings of initial doubling of the tracheal circumference with slide tracheoplasty, several children showed no change or even a decrease in airway size after slide tracheoplasty. Although swelling and granulation tissue may have contributed to this, too, the retrospective nature of the study made it difficult to confirm that documentation of sizing at the time of slide tracheoplasty always referred to size at the level of the complete rings and prior to performing the slide tracheoplasty. These factors may have affected this calculation.

This study has several limitations given the relatively small population and retrospective nature; however, with...
Figure 4. Depiction of airway growth over time postslide tracheoplasty. Each line represents a different patient, with each point marking the size at a follow-up microlaryngoscopy and bronchoscopy.

Table 3. Timing of Airway Evaluations and Sizing.\textsuperscript{a}

<table>
<thead>
<tr>
<th>MLB Information</th>
<th>Age and Time, mo</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age at slide tracheoplasty</td>
<td>7 (3, 14.25)</td>
</tr>
<tr>
<td>Duration of follow-up</td>
<td>57 (40.25, 96)</td>
</tr>
<tr>
<td>Number of MLBs per patient (n = 426)</td>
<td>10 (7, 12.75)</td>
</tr>
<tr>
<td>Time interval between MLBs</td>
<td>3 (0, 11)</td>
</tr>
<tr>
<td>Number of MLBs with sizing per patient (n = 257)</td>
<td>6 (4, 8)</td>
</tr>
<tr>
<td>Time interval between sizing</td>
<td>5 (0, 13)</td>
</tr>
</tbody>
</table>

Abbreviations: IQR, interquartile range; MLB, microlaryngoscopy and bronchoscopy. \textsuperscript{a}Values presented are median (IQR: 25, 75%) and range. Time intervals are in months.

Table 4. Adjunctive Airway Surgeries Performed Throughout Postoperative Follow-up.

<table>
<thead>
<tr>
<th>Adjunctive Surgeries</th>
<th>No. of Patients Requiring</th>
<th>No. of Procedures Performed</th>
</tr>
</thead>
<tbody>
<tr>
<td>Balloon dilation\textsuperscript{a}</td>
<td>8</td>
<td>20</td>
</tr>
<tr>
<td>Tracheostomy</td>
<td>3</td>
<td>2</td>
</tr>
<tr>
<td>Removal of granulation</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>Supraglottoplasty</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>ssLTP w/ACCG</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>Palmaz stent placement</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>Endoscopic VF lateralization</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>Adenotonsillar surgery</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>Endoscopic cleft repair</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>Bugbee cautery of pig bronchus</td>
<td>1</td>
<td>3</td>
</tr>
<tr>
<td>Stoma revision</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Revision slide tracheoplasty\textsuperscript{b}</td>
<td>1</td>
<td>1</td>
</tr>
</tbody>
</table>

Abbreviations: ssLTP w/ACCG, single-stage laryngotraceoplasty with anterior costal cartilage graft; VF, vocal fold. \textsuperscript{a}Balloon dilation was performed a median of 2 times on each of the 8 children with the first at a median of 1 month after slide tracheoplasty. \textsuperscript{b}Revision slide tracheoplasty was required in 1 patient 66 months after the initial procedure.
complete tracheal rings being a rare phenomenon, this is the largest series of patients with repaired complete tracheal rings and reports longitudinal data. As this was a retrospective study, accurate sizing information was not available for each MLB performed. In addition, sizing was not performed at some MLBs due to concern for exacerbating the disease process, potential to interfere with healing postoperatively, or if the airway size was obviously adequate. This, in all likelihood, causes an underestimation of change in airway diameter initially because the airway is often not sized if it is visibly adequate and the child is asymptomatic. Another limitation is that many children are referred to our institution for evaluation of complete tracheal rings. As a result, several of the patients had follow-up at outside institutions before and/or after evaluation at our institution, making some follow-up information unavailable to us. The individual patterns of change in airway size depicted in Figure 4 are certainly influenced by the frequency of the endoscopies and sizing; however, the overall growth rates are calculated as a function of time since surgery to minimize the influence of this variability. It is unlikely that children underwent major interventions after slide tracheoplasty at outside institutions after being released from our care given the specialized nature of the postslide trachea; however, that information may not be available to us. Children who underwent surgical interventions prior to evaluation at our institution were excluded to help minimize confounding factors.

Our institution’s current practice has been to follow these children with serial endoscopy through puberty at increasingly longer intervals between procedures. However, this study is reassuring that perhaps fewer endoscopies for a shorter duration of follow-up could be reasonable in asymptomatic children while reducing repetitive anesthetic exposures. This does necessitate, however, having informed, reliable patients who will be able to recognize issues and present accordingly. Future studies directed at determining the ideal point (ie, airway size or duration out from surgery) that can be considered “safe” to stop surveillance endoscopy, as well as ways to predict patients who are likely to not have growth over time and/or to develop symptoms in the long term, would be useful.

Conclusion

Slide tracheoplasty has become the preferred method of surgical management of complete tracheal rings due to the minimization of complications that plague older methods of treating complete tracheal rings. This is the first and largest study documenting longitudinal growth of RCTRs after slide tracheoplasty. Children with RCTRs require close monitoring in the early postoperative period to ensure adequate healing and recovery. After initial healing, the airway typically grows at a relatively normal rate, and frequent endoscopy may not be necessary. The interval between MLBs can be increased as the child gets older, as the airway increases in size, and as long as symptoms are minimal.

Author Contributions

Lyndy J. Wilcox, conception/design, acquisition/analysis and interpretation of data, drafting and revising manuscript; Claudia Schweiger, conception/design, acquisition/analysis and interpretation of data, drafting and revising manuscript; Catherine K. Hart, conception/design, interpretation of data, drafting and revising manuscript; Alessandro de Alarcon, conception/design, interpretation of data, revising of manuscript; Nithin S. Peddireddy, conception/design, acquisition of data, revising of manuscript; Michael J. Rutter, conception/design, analysis and interpretation of data, revising of manuscript.

Disclosures

Competing interests: Michael J. Rutter, consultant/patent holder: Bryan Medical (Aeris balloon dilator); consultant: Tracoe; consultant (no financial relationship): Boston Medical Products (suprasto-mal stent).

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References


