Descending Aortopexy and Posterior Tracheopexy for Severe Tracheomalacia and Left Mainstem Bronchomalacia

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Posterior descending aortopexy can relieve posterior intrusion of the left mainstem bronchus that may limit the effectiveness of posterior tracheobronchopexy. We review outcomes of patients undergoing both descending aortopexy and posterior tracheopexy for severe tracheobronchomalacia with posterior intrusion and left mainstem compression to determine if there were resolution of clinical symptoms and bronchoscopic evidence of improvement in airway collapse. All patients who underwent both descending aortopexy and posterior tracheopexy from October 2012 to October 2016 were retrospectively reviewed. Clinical symptoms, tracheomalacia scores based on standardized dynamic airway evaluation by anatomical region, and persistent airway intrusion requiring reoperation were collected. Data were analyzed by Wald and Wilcoxon signed-rank tests. Thirty-two patients underwent descending aortopexy and posterior tracheopexy at median age of 18 months (interquartile range 6-40 months). Median follow-up was 3 months (interquartile range 1-7 months). There were statistically significant improvements in clinical symptoms postoperatively, including cough, noisy breathing, prolonged and recurrent respiratory infections, ventilator dependence, blue spells, and brief resolved unexplained events (all $P < 0.001$), as well as exercise intolerance ($P = 0.033$), transient respiratory distress requiring positive pressure ($P = 0.003$), and oxygen dependence ($P = 0.007$). Total tracheomalacia scores improved significantly ($P < 0.001$), with significant segmental improvements in the middle ($P = 0.003$) and lower ($P < 0.001$) trachea, and right ($P = 0.011$) and left ($P < 0.001$) mainstem bronchi. Two patients (6%) had persistent airway intrusion requiring reoperation with anterior aortopexy or tracheopexy. Descending aortopexy and posterior tracheopexy are effective in treating severe tracheobronchomalacia and left mainstem intrusion with significant improvements in clinical symptoms and degree of airway collapse on bronchoscopy.

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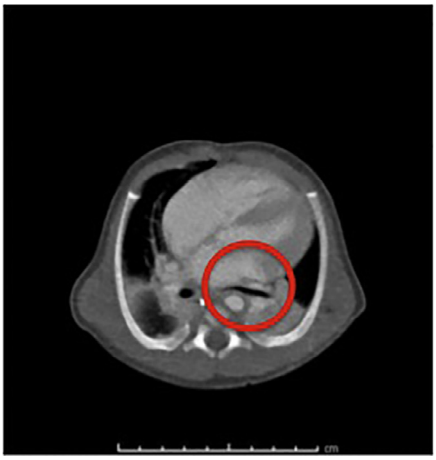

Cardiomegaly and a left descending aorta compressing the left mainstem bronchus.

Central Message

Descending aortopexy and posterior tracheopexy are clinically effective in treating severe tracheobronchomalacia and left mainstem intrusion with significant symptom and anatomical improvements.

Perspective Statement

Descending aortopexy and posterior tracheopexy are effective in treating severe tracheobronchomalacia and left mainstem intrusion with significant improvements in clinical symptoms and degree of airway collapse on bronchoscopy. Complex cases warrant an individualized and flexible surgical approach guided by preoperative imaging and intraoperative bronchoscopy in multidisciplinary specialized centers.

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INTRODUCTION

Tracheobronchomalacia refers to a weakness or deformation of the airway such that it is more susceptible to collapse with changes in pressure and compression by adjacent thoracic structures.[1] It is often associated with esophageal atresia (EA), tracheoesophageal fistula (TEF), and congenital heart disease (CHD).[2] Severe tracheobronchomalacia is characterized by dynamic airway collapse in spontaneously breathing patients with anterior vascular compression and posterior membranous tracheal intrusion.[3] Anterior ascending aortopexy addresses anterior vascular compression by indirectly elevating the anterior wall of the trachea but does not directly address posterior membranous tracheal intrusion.[4] We recently reported a series of patients who underwent posterior tracheopexy for severe symptomatic tracheomalacia with posterior intrusion with promising short-term results.[5] The effectiveness of posterior tracheopexy can be limited by left mainstem bronchomalacia in some patients. Posterior descending aortopexy can be used to relieve left mainstem posterior intrusion and compression between the descending aorta and the pulmonary artery. We now review a series of patients who underwent both descending aortopexy and posterior tracheopexy for severe symptomatic tracheobronchomalacia with posterior intrusion and left mainstem compression to determine if there were resolution of clinical symptoms and bronchoscopic evidence of improvement in airway collapse.

METHODS

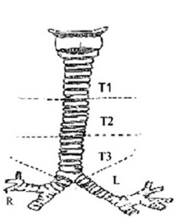
The Esophageal and Airway Treatment (EAT) Center at Boston Children's Hospital is a multidisciplinary care team consisting of 3 pediatric surgeons, 1 pediatric cardiothoracic surgeon, 1 pediatric pulmonologist, and 2 pediatric gastroenterologists. We retrospectively reviewed all patients who underwent both descending aortopexy and posterior tracheopexy at Boston Children's Hospital from October 2012 to October 2016 under an approved institutional review board protocol (IRB-P00021702).

Patient demographics, pre- and postoperative clinical symptoms and airway evaluation, surgical techniques, and persistent airway intrusion requiring reoperation were collected. Clinical symptoms included cough, barking cough, noisy breathing, prolonged pulmonary infection, recurrent pulmonary infections, exercise intolerance, transient respiratory distress requiring positive pressure, oxygen dependence, ventilator dependence, blue spells, and brief resolved unexplained events (BRUEs) (formerly known as apparent life-threatening events).

Pre- and postoperative endoscopic airway evaluation was performed by the primary surgeons involved. Diagnostic laryngoscopy and bronchoscopy was done under general anesthesia in spontaneously breathing patients. After assessing supraglottic structures and vocal cord function, the vocal cords were anesthetized with topical lidocaine, the larynx was assessed for presence of a laryngeal cleft, and a Hopkins II rod lens was inserted through the cords to assess for TEF, tracheal diverticulum, and dynamic motion in the tracheobronchial tree throughout the respiratory cycle. A standardized tracheomalacia scoring system based on dynamic airway evaluation was used to determine pre- and postoperative tracheomalacia scores (Table).[3,5,6] The tracheobronchial tree was scored by the percentage of open airway during cough or Valsalva out of 100 at each anatomical region: upper (T1), middle (T2), and lower (T3) trachea, and right and left mainstem bronchi, with a maximum score of 500. Dynamic airway multidetector computed tomography (MDCT) was performed preoperatively to evaluate aberrant vascular anatomy or associated lung parenchymal disease, and to identify the artery of Adamkiewicz.[6] As it is difficult to justify postoperative radiation and routine MDCT imaging in asymptomatic children, bronchoscopic airway evaluation was used for postoperative follow-up.

The operating surgeon determined the operative plan and approach based on endoscopic evaluation and preoperative MDCT, as part of a multidisciplinary team. Generally, patients with associated esophageal disease underwent right posterior thoracotomy, those with cardiac disease underwent median sternotomy, and those with vascular rings underwent left thoracotomy. The esophagus, back wall of the trachea, thoracic duct, and aorta were fully dissected and mobilized, taking care to protect the left vagus nerve and left recurrent laryngeal nerve. In patients undergoing sternotomy, the ductal ligament was often divided to extensively mobilize the ascending aorta, transverse aortic arch, and descending aorta. A recurrent TEF or residual tracheal diverticulum from a previously repaired TEF was corrected if present by resecting the TEF or diverticulum flush with the tracheal wall under endoscopic visualization. Pexy procedures were all done under direct flexible

Table. Tracheomalacia Scores



Location	Preoperative (n = 32)	Postoperative (n = 15)	*P* Value
T1	80 (70-95)	100 (80-100)	0.182
T2	0 (0-45)	75 (60-100)	0.003*
T3	0 (0-30)	100 (70-100)	<0.001*
Right bronchus	78 (12-100)	100 (100-100)	0.011*
Left bronchus	20 (0-50)	70 (50-100)	<0.001*
Total	215 (145-268)	450 (360-475)	<0.001*

Pre- and postoperative tracheomalacia scores based on standardized bronchoscopic evaluation. Scores are percentage of open airway out of 100 for each anatomical region. Data are median (IQR).
*Statistically significant.

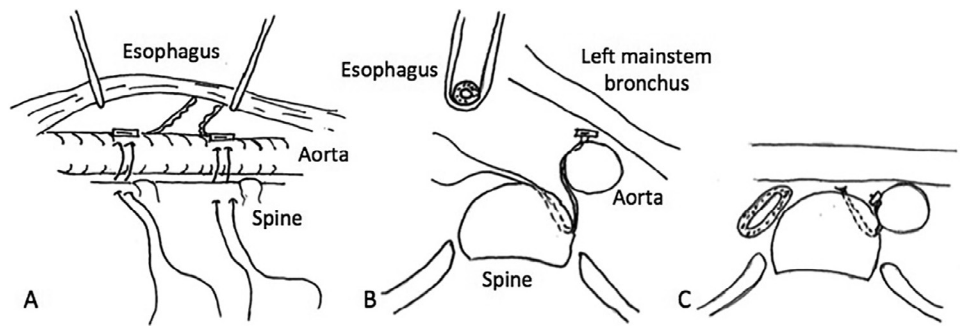


Figure 1. Posterior descending aortopexy. (A) Anatomical relationship of aorta, esophagus, and spine. (B) Cross-sectional view, in which the esophagus is rotated to the right and the descending aorta is moved to the left and secured to the side of the spine as posteriorly as necessary to relieve posterior pressure off the left mainstem bronchus. (C) Descending aortopexy sutures are tied, relieving left mainstem posterior intrusion and compression between the descending aorta and the pulmonary artery.

bronchoscopic guidance. Posterior descending aortopexy was performed by passing autologous pleural or pericardial pledgeted polypropylene sutures to secure the aorta to the side of the spine, and as posteriorly as necessary to relieve posterior pressure off the left mainstem bronchus (Fig. 1). This posterior movement of the aorta may necessitate dividing 1 or more intercostals, and preoperative MDCT helps to ensure that the artery of Adamkiewicz is not divided. Arm-leg blood pressure measurements or arterial line tracing monitoring was used to confirm no descending aortic gradient. Posterior tracheopexy was performed by passing autologous pleural or pericardial pledgeted sutures into but not through the posterior tracheal membrane, and securing them to the anterior longitudinal spinal ligament, in such a fashion as to optimize tracheal lumen and posterior tracheal membrane support. Suture placement is guided by intraoperative bronchoscopic guidance to avoid full thickness bites and to optimize placement of the sutures. Additional airway pexy procedures were similarly performed by passing pledgeted sutures to secure the bronchial posterior membrane posteriorly to the lateral edges of the anterior longitudinal ligament of the spine. In patients undergoing sternotomy, anterior pexy sutures to support and displace the vasculature and airway were then passed through the sternum and secured under direct bronchoscopic visualization after sternal closure.

Statistical Analysis

To assess resolution of clinical symptoms, the percentage of patients with each symptom pre- and postoperatively was compared by the Wald chi-square test using logistic regression modeling with a generalized estimating equations approach to account for the binary paired data.[7] Changes in tracheomalacia scores for each airway segment were determined by the Wilcoxon signed-ranks test.[8] Freedom from reoperation was estimated using the Kaplan-Meier product-limit method.[9] Statistical analysis was performed using IBM SPSS Statistics (version 23.0, IBM Corporation, Armonk, NY). A two-tailed $P < 0.05$ was considered statistically significant.

RESULTS

Thirty-two patients underwent descending aortopexy and posterior tracheopexy at median age of 18 months (interquartile range [IQR] 6-40 months). Sixty-three percent (20 patients) were male. Median estimated gestational age was 34 weeks (IQR 31-36 weeks). Sixty-six percent (21 patients) were associated with EA, including 18 patients with type C EA, 2 patients with type A EA, and 1 patient with type B EA. Nineteen percent (6 patients) had long gap EA. Sixty-nine percent (22 patients) had associated cardiac disease, and 19% (6 patients) had VACTERL syndrome. Sixty-three percent (20 patients) had a prior EA repair, and 19% (6 patients) had a prior anterior aortopexy. Nineteen percent (6 patients) had vascular rings, including 3 patients with double aortic arch, 2 patients with right aortic arch with Kommerell diverticulum, and 1 patient with circumflex aorta. Nine percent (3 patients) had a prior vascular ring division, 3% (1 patient) had a prior coarctation repair, and 13% (4 patients) had a prior patent ductus arteriosus ligation. Three percent (1 patient) had double superior vena cava and 3% (1 patient) had dysplastic pulmonary valve stenosis. Six percent (2 patients) had tetralogy of Fallot, one of which had undergone prior repair. Twenty-two percent (7 patients) had atrial and ventricular septal defects, 3 of which had undergone prior septal defect closures.

Upper airway anomalies were common on preoperative and intraoperative bronchoscopy. Three percent (1 patient) had laryngomalacia. No patients had preoperative vocal cord dysfunction or paralysis. Twenty-two percent (7 patients) had laryngeal clefts, 6 patients with type 1 clefts and 1 patient with a type 3 cleft. Nineteen percent (6 patients) had some degree of subglottic stenosis. The tracheobronchial tree was scored by anatomical region (Table). Preoperatively, the middle (T2) trachea, lower (T3) trachea, and left mainstem bronchus were the most severely affected, with median scores of 0 (IQR 0-45), 0 (IQR 0-30), and 20 (IQR 0-50), respectively.

Operative approach was by right thoracotomy in 56% (18 patients), median sternotomy in 22% (7 patients), left thoracotomy in 19% (6 patients), and right neck dissection with right thoracotomy in 3% (1 patient). All patients underwent descending aortopexy and posterior tracheopexy under intraoperative bronchoscopic guidance. Fifty-three percent (17 patients) underwent additional airway or vascular pexy procedures, including left mainstem bronchopexy in 38% (12 patients), right mainstem bronchopexy in 16% (5 patients), innominate artery pexy in 9% (3 patients), anterior tracheopexy in 9% (3 patients), and anterior aortopexy in 9% (3 patients). Six percent (2 patients) had an aberrant subclavian artery behind the trachea, requiring mobilization of the artery in 1 patient and division of the artery in 1 patient. Fifty-three percent (17 patients) had an associated tracheal diverticulum that was resected flush with the trachea. Concomitant procedures included septal defect closure in 13% (4 patients), vascular ring division in 9% (3 patients), tetralogy of Fallot repair in 3% (1 patient), and pulmonary valve replacement in 3% (1 patient).

Median days on the ventilator after surgery were 4 days (IQR 0-11 days). Median total intensive care unit stay was 10 days (IQR 2-23 days). Median total hospital length of stay was 15 days (IQR 7-42 days). There were no significant early complications including hemorrhage or infection. There were no complications with erosion of the pledgetted sutures into the aorta or trachea, as the pledgets are autologous. There were no mortalities.

Median clinical follow-up was 3 months (IQR 1-7 months). There were statistically significant improvements in clinical symptoms postoperatively, including cough, barking cough, noisy breathing, prolonged and recurrent respiratory infections, ventilator dependence, blue spells, and BRUEs (all $P < 0.001$), as well as exercise intolerance ($P = 0.033$), transient respiratory distress requiring positive pressure ($P = 0.003$), and oxygen dependence ($P = 0.007$) (Fig. 2). At latest follow-up, no patients had recurrence of a blue spell or BRUE.

Forty-seven percent (15 patients) underwent postoperative follow-up evaluation with bronchoscopy at median of 2 months (IQR 1-6.5 months). Total tracheomalacia scores on bronchoscopy improved significantly ($P < 0.001$), with significant segmental improvements in the middle (T2) ($P = 0.003$) and lower (T3) ($P < 0.001$) trachea, and the right ($P = 0.011$) and left ($P < 0.001$) mainstem bronchi (Table). The greatest areas of numerical improvement were in the segments most affected preoperatively, namely, the middle (T2) and lower (T3) trachea, as well as the left mainstem bronchus.

Six percent (2 patients) had persistent airway intrusion requiring reoperation, using anterior aortopexy and anterior tracheopexy. The 2 reoperations were within 2 months after the index surgery. Kaplan-Meier analysis estimated 92% of patients to be free from reoperation at 3 months follow-up (95% confidence interval 85%-99%).

Six percent (2 patients) had tracheostomies preoperatively, all for severe tracheomalacia. Overall, 9% (3 patients) had tracheostomies postoperatively, with no significant difference compared with preoperatively ($P = 0.31$). The 2 patients with preoperative tracheostomies on the ventilator were able to wean to tracheostomy collar postoperatively. One additional patient with associated severe cardiac disease, transferred intubated on mechanical ventilation, had a tracheostomy placed for prolonged intubation and ventilator weaning.

DISCUSSION

Tracheobronchomalacia is an underestimated disease, given the wide spectrum of disease with nonspecific chronic respiratory symptoms that are commonly misdiagnosed.[1,2,10,11] It is a common respiratory problem in patients with EA and CHD.[12-15] Among at least patients with CHD, tracheobronchomalacia has been

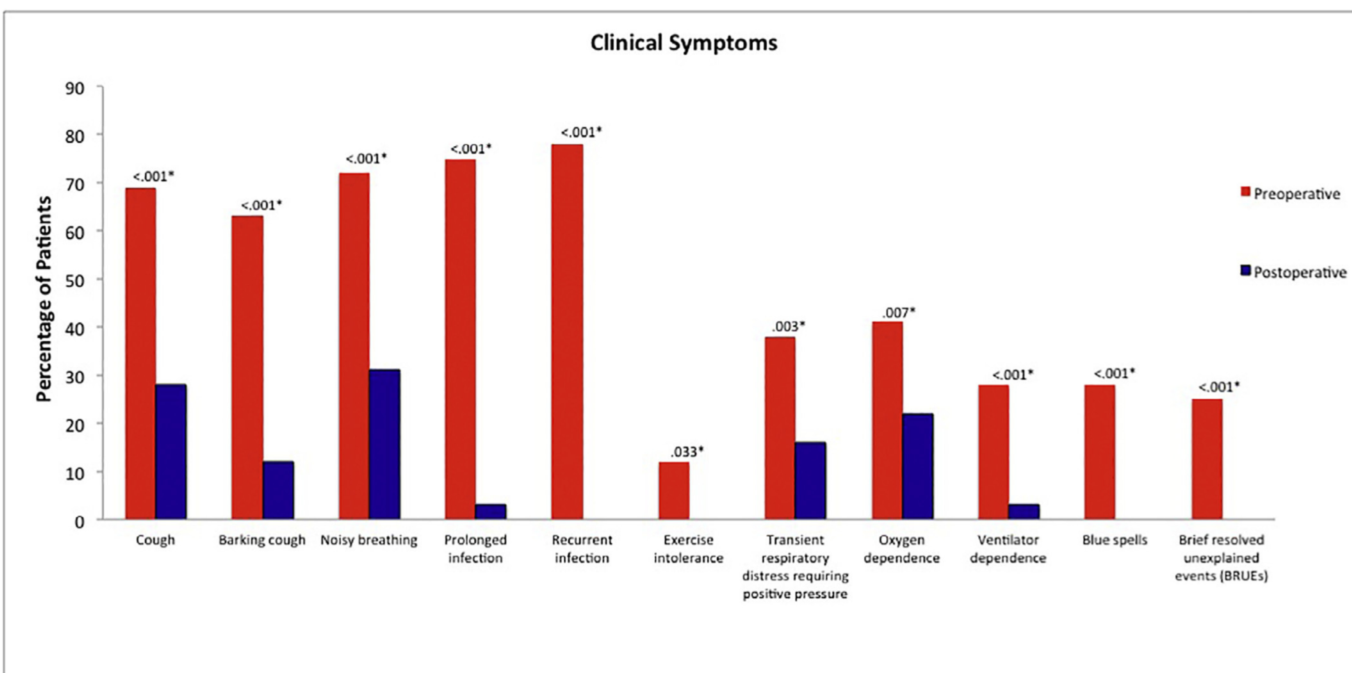


Figure 2. Pre- and postoperative clinical symptoms. (Color version of figure is available online.)

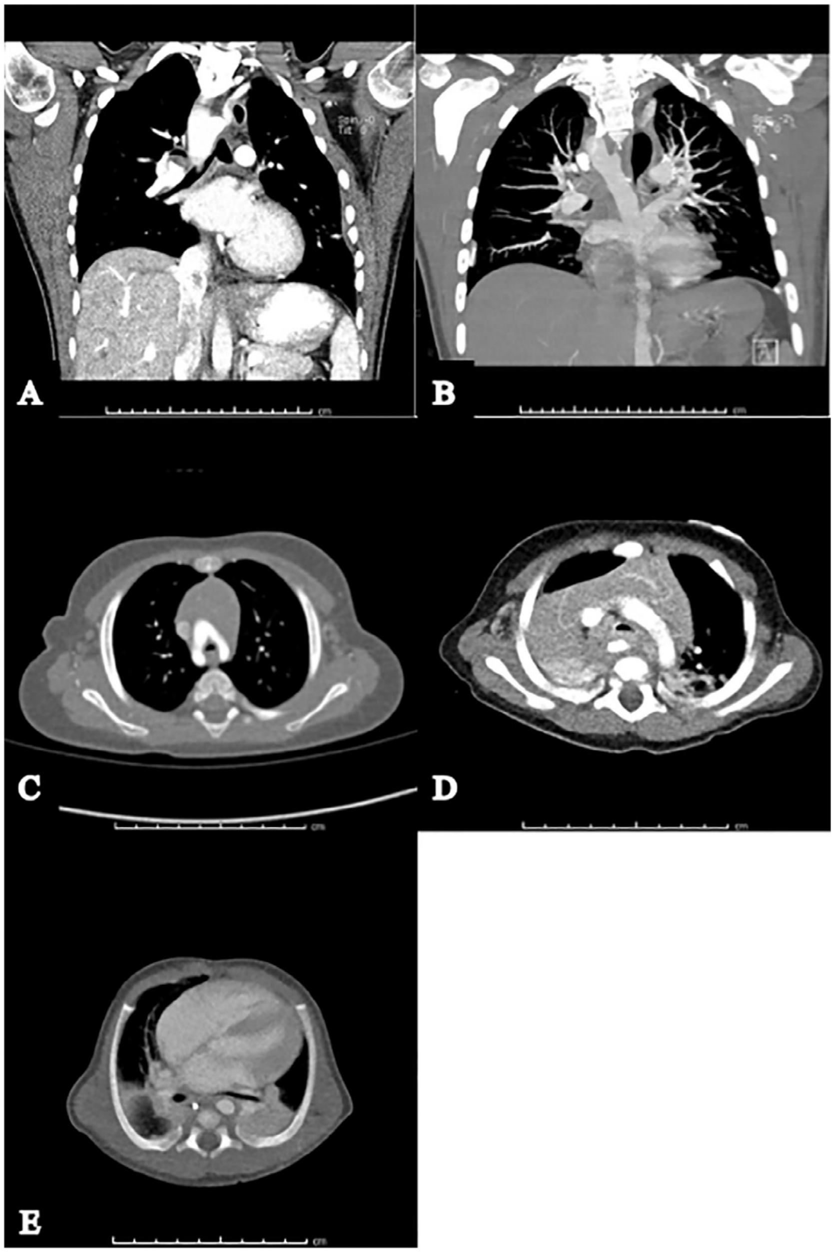


Figure 3. Preoperative dynamic airway MDCT of complex cases of tracheobronchomalacia. (A) Right aortic arch with large Kommerell diverticulum. (B) Right to left circumflex aorta. (C) Double aortic arch compressing the trachea. (D) Left aortic arch compressing the trachea. (E) Left descending aorta compressing the left mainstem bronchus.

associated with increased ventilator days, length of stay, and mortality.[15-17] Excessive airway collapse leads to ineffective ventilation and poor clearance of secretions, resulting in frequent respiratory infections, respiratory failure, and apneic events, all of which are less well tolerated in patients with EA and CHD, underscoring the importance of early diagnosis in these populations.[1,2,10,18] Our multidisciplinary care team routinely uses a standardized reporting and scoring system based on anatomical region for endoscopic evaluation.[3,5,6] The greater the severity of airway collapse, indicated by a lower tracheomalacia score, combined with the presence of clinical symptoms, may indicate the need for surgical intervention, possibly concomitant with EA or CHD repair.

Preoperative dynamic airway MDCT is used in conjunction with bronchoscopy to inform the operative plan. MDCT is particularly useful in evaluating complex cases with aberrant vascular anatomy and associated cardiac or esophageal anomalies. Several different types of disease resulting in complex tracheobronchomalacia include a right aortic arch with large Kommerell diverticulum (Fig. 3A), a right to left circumflex aorta (Fig. 3B), a double aortic arch compressing the trachea (Fig. 3C), a left aortic arch compressing the trachea (Fig. 3D), and a left descending aorta compressing the left mainstem bronchus (Fig. 3E).

The management of severe tracheobronchomalacia remains difficult with little consensus on treatment and surgical approach.[1,2,15,19] Surgical options include pexy procedures (ascending and descending aortopexy, anterior and posterior tracheopexy), tracheal resection, internal stents, and external stabilization.[1-5,15,20-24] Anterior ascending aortopexy indirectly supports the anterior tracheal wall and is the most commonly used technique, but has a reported failure rate of 10%-25% in the literature.[15,22-24] Direct anterior and posterior tracheopexy, as first reported by our group, improve airway patency by directly addressing anterior malformed tracheal cartilage and posterior membranous tracheal intrusion.[2,3,5]

Left mainstem bronchomalacia is a challenging entity that may limit airflow, mucus clearance, and the effectiveness of direct tracheopexy in some cases, which is the focus of this paper.[25] The descending aorta may be anteriorly displaced, intruding into the back wall of the left mainstem bronchus, whereas the pulmonary artery may be posteriorly displaced, compressing the airway from the front, resulting in left mainstem vascular compression. Surgical options include posterior descending aortopexy, descending aortic translocation, pulmonary artery anterior fixation, internal stents, and external splints.[26-32]

In this series, we show that descending aortopexy and posterior tracheopexy are clinically effective in treating severe tracheobronchomalacia with posterior intrusion and left mainstem compression. Postoperatively, there were significant improvements in clinical symptoms, as well as anatomical tracheomalacia scores. Anterior and posterior tracheopexy or bronchopexy provide direct support to the airway, whereas anterior ascending and posterior descending aortopexy indirectly support the airway by directly addressing vascular compression of the airway. Complex cases of severe tracheobronchomalacia, especially those with altered anatomy associated with EA or CHD, warrant an individualized and flexible surgical approach guided by intraoperative bronchoscopy. In fact, in our series, 53% of patients underwent concomitant airway and vascular pexy procedures to optimize each individual airway.

Posterior descending aortopexy is a technically challenging procedure. Through a right posterior thoracotomy approach, sutures are placed in the anterior aspect of the descending aorta and then blindly placed on the left side of the spine close to the costovertebral junction by passing the needle out the anterior longitudinal ligament of the spine. Through a median sternotomy approach, it can be even more challenging to mobilize and expose the descending aorta and place sutures to posteriorly displace it. We find it best to monitor upper and lower extremity blood pressure or arterial line tracings to avoid aortic stenosis.

There are a number of limitations to this study. Retrospective chart review was used to collect the data and follow-up, including pre- and postoperative clinical symptoms and bronchoscopy findings. Although patients are followed up closely by our multidisciplinary clinic, further studies could utilize a prospective structured clinical symptom questionnaire to further standardize reporting. Bronchoscopy can be subjective and was performed by 3 primary operating surgeons. One study in adults showed appropriate inter- and intraobserver reliability in flexible bronchoscopy; however, less is known in the pediatric population.[33] Future work can include bronchoscopic analysis by independent observers to make a more statistically valid comparison. Postoperative endoscopic evaluation was not available for all patients; however, we used the standardized scoring system to demonstrate resolution of tracheomalacia postoperatively in those evaluated. Our standard protocol for endoscopic postoperative evaluation is at 1 year for longitudinal airway assessment if clinically asymptomatic, unless the patient is undergoing another scheduled procedure. Our study cohort included a heterogeneous group of complex patients often requiring concomitant airway or vascular pexy procedures or adjunct therapies that may have contributed to outcomes and confounded the influence of surgical treatment alone. As our preference is to correct all anomalies that may affect airway or cardiac function at the initial operation to try to prevent multiple reoperations, it would be nearly impossible to isolate patients undergoing only 1 procedure. Follow-up intervals were relatively short term and variable.

In conclusion, descending aortopexy and posterior tracheopexy are effective in treating severe tracheobronchomalacia and left mainstem compression with significant improvement or resolution of clinical symptoms and degree of airway collapse on bronchoscopy. Further studies to follow long-term outcomes of this technique are certainly warranted and are ongoing. Given the heterogeneity and complexity of this patient population with significant morbidity, treatment and long-term follow-up is best done in multidisciplinary specialized centers for individualized patient care.

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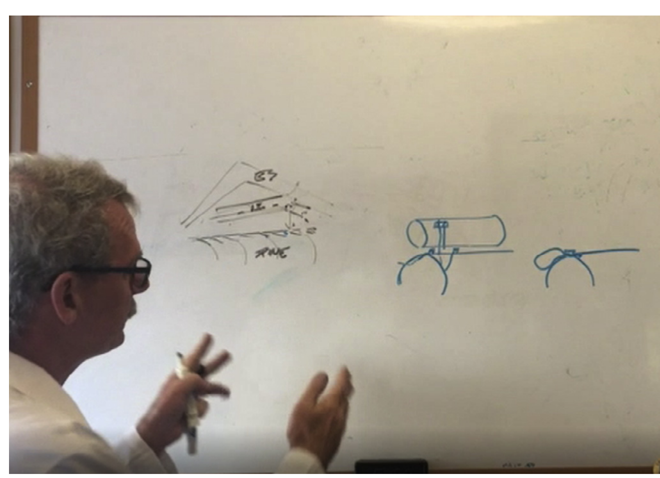
SUPPLEMENTARY MATERIAL

The following is the supplementary data to this article:



Video 1. Descending aortopexy and posterior tracheopexy. Operative procedure and its relevance as discussed by Dr Russell Jennings.

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