Journal of Pediatric Surgery 52 (2017) 951–955

Contents lists available at ScienceDirect

Journal of Pediatric Surgery

journal homepage: www.elsevier.com/locate/jpedsurg

Posterior tracheopexy for severe tracheomalacia

Hester F. Shieh [a], C. Jason Smithers [a], Thomas E. Hamilton [a], David Zurakowski [a], Lawrence M. Rhein [b], Michael A. Manfredi [c], Christopher W. Baird [d], Russell W. Jennings [a,*]

[a] Department of Surgery, Boston Children's Hospital, Harvard Medical School, Boston, MA 02115, United States
[b] Department of Pulmonology, Boston Children's Hospital, Harvard Medical School, Boston, MA 02115, United States
[c] Department of Gastroenterology, Boston Children's Hospital, Harvard Medical School, Boston, MA 02115, United States
[d] Department of Cardiovascular Surgery, Boston Children's Hospital, Harvard Medical School, Boston, MA 02115, United States

ARTICLE INFO

Article history:
Received 2 March 2017
Accepted 9 March 2017

Key words:
Posterior tracheopexy
Tracheomalacia
Aortopexy

ABSTRACT

Purpose: In severe tracheomalacia, aortopexy addresses anterior vascular compression, but does not directly address posterior membranous tracheal intrusion. We review patient outcomes of posterior tracheopexy for tracheomalacia with posterior intrusion to determine if there were resolution of clinical symptoms and bronchoscopic evidence of improvement in airway collapse.

Methods: All patients who underwent posterior tracheopexy from October 2012 to March 2016 were retrospectively reviewed. Clinical symptoms, tracheomalacia scores based on standardized dynamic airway evaluation by anatomical region, and persistent airway intrusion were collected. Data were analyzed by Wald and Wilcoxon signed-ranks tests.

Results: 98 patients (51% male) underwent posterior tracheopexy at a median age of 15 months (IQR 6–33 months). Median follow-up was 5 months (range 0.25–36 months). There were statistically significant improvements in clinical symptoms postoperatively, including cough, noisy breathing, prolonged and recurrent respiratory infections, transient respiratory distress requiring positive pressure, oxygen dependence, blue spells, and apparent life-threatening events ($p < 0.001$), as well as ventilator dependence ($p = 0.04$). Tracheomalacia scores on bronchoscopy improved significantly in all regions of the trachea and bronchi ($p < 0.001$). 9.2% had persistent airway intrusion requiring reoperation, usually with aortopexy.

Conclusions: Posterior tracheopexy is effective in treating severe tracheomalacia with significant improvements in clinical symptoms and degree of airway collapse on bronchoscopy.

Level of evidence: Level III, treatment study.

Tracheomalacia refers to a weakness of the trachea such that the airway is more susceptible to collapse with changes in pressure and compression by adjacent thoracic structures. Severe tracheomalacia is characterized by coaptation of the airways with anterior and posterior collapse during expiration in spontaneously breathing patients. Tracheomalacia is often associated with esophageal atresia (EA), tracheoesophageal fistula (TEF), and congenital heart disease (CHD) [1–3]. Aortopexy addresses the anterior vascular compression component by indirectly elevating the anterior wall of the trachea, but does not directly address posterior membranous tracheal intrusion. We previously reported a small series of patients who underwent direct anterior and/or posterior tracheopexy for severe tracheomalacia with promising short-term results [1]. We now review a larger series of patients who primarily underwent posterior tracheopexy for severe symptomatic tracheomalacia with posterior intrusion to determine if there was resolution of clinical symptoms. We present our standardized tracheomalacia scoring system based on dynamic airway evaluation to determine if there was bronchoscopic and clinical evidence of improvement in airway collapse.

1. Methods

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

Patient demographics, preoperative 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

* Corresponding author at: Department of Surgery, Boston Children's Hospital, Harvard Medical School, 300 Longwood Ave., Fegan 3, Boston, MA 02115, United States. Tel.: +1 617 355 3038; fax: +1 617 730 0477.
E-mail address: russell.jennings@childrens.harvard.edu (R.W. Jennings).

http://dx.doi.org/10.1016/j.jpedsurg.2017.03.018
0022-3468/

dependence, ventilator dependence, blue spells, and apparent life-threatening events (ALTEs).

Preoperative and postoperative endoscopic airway evaluation was performed by the primary surgeons involved. Diagnostic laryngoscopy/bronchoscopy (DLB) 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. To determine preoperative and postoperative tracheomalacia scores, the tracheobronchial tree was evaluated by anatomical region and percentage of open airway with a modified standardized scoring system (Table 1) [1,4]. Anatomic regions were classified into upper (T1), middle (T2), and lower (T3) trachea, as well as right and left mainstem bronchi. The degree of open airway at each of the five anatomic regions was scored out of 100 with a maximum score of 500. Additional dynamic airway multidetector computed tomography (MDCT) was performed at the discretion of the operating surgeons to evaluate for aberrant vascular anatomy or associated lung parenchymal disease [4].

The operating surgeon determined the operative plan and approach based on endoscopic evaluation and preoperative MDCT if available. Generally patients with associated esophageal disease underwent right posterior thoracotomy, whereas those with cardiac disease underwent sternotomy. The esophagus, back wall of the trachea, thoracic duct, and/or 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 fully 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 bronchoscopic visualization. Posterior tracheopexy was performed by passing autologous pledgeted polypropylene sutures into but not through the posterior tracheal membrane, and securing them to the anterior longitudinal spinal ligament under direct bronchoscopic guidance. Aortopexy and anterior tracheopexy were performed to elevate the anterior wall of the trachea, when necessary. Anterior tracheopexy was performed by passing pledgeted sutures through the rings under direct endoscopic visualization. In patients undergoing sternotomy, these sutures were then passed through the sternum and secured under direct bronchoscopic visualization following sternal closure. In patients with multiple disease processes, surgeons favored addressing all concerns during one operative case when possible in an attempt to avoid reoperations.

To assess resolution of clinical symptoms, the percentage of patients with each symptom preoperatively and postoperatively was compared by the Wald chi-square test using logistic regression modeling with a generalized estimating equations (GEE) approach to account for the binary paired data. Changes in tracheomalacia scores for each airway segment were determined by the Wilcoxon signed-ranks test. Statistical analysis was performed using IBM SPSS Statistics (version 23.0, IBM, Armonk, NY). A two-tailed p value <0.05 was considered statistically significant.

2. Results

98 patients underwent posterior tracheopexy at a median age of 15 months (interquartile range (IQR) 6–33 months). 50 patients (51.0%) were male. Median estimated gestational age (EGA) was 35 weeks (IQR 33–38 weeks). 86 patients (87.8%) had an associated EA with or without TEF, and 30 patients (30.6%) had long gap EA. 35 patients (35.7%) had associated CHD, and 33 patients (33.7%) had VACTERL syndrome. 68 patients (69.4%) had a prior EA repair and 8 patients (8.2%) had a prior aortopexy.

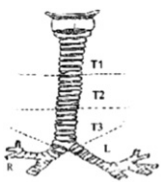
All patients had preoperative and intraoperative bronchoscopy. On upper airway evaluation, 3 patients (3.1%) had laryngomalacia and 4 patients (4.1%) had preoperative left vocal cord paralysis. 20 patients (20.4%) had laryngeal clefts, 10 with type 1 clefts, 6 with type 2 clefts, 3 with type 3 clefts, and 1 with a type 4 cleft. 11 patients (11.2%) had some degree of subglottic stenosis. The tracheobronchial tree was scored by anatomic region (Table 1). The middle (T2) and lower (T3) trachea were the most severely affected preoperatively, with median scores of 5 (IQR 0–30) and 5 (IQR 0–25), respectively.

Operative approach was by right thoracotomy in 79 patients (80.6%) and median sternotomy in 11 patients (11.2%). Other operative approaches included left thoracotomy in 3 patients (3.1%), left neck dissection in 1 patient (1.0%), and combined approaches in the remaining 4 patients (4.1%). All patients underwent posterior tracheopexy under intraoperative bronchoscopic guidance. 18 patients (18.4%) underwent additional procedures to open their airway, including aortopexy in 11 patients (11.2%), left mainstem bronchopexy in 6 patients (6.1%), anterior tracheopexy in 5 patients (5.1%), pulmonary artery pexy in 1 patient (1.0%), and innominate artery pexy in 1 patient (1.0%). 5 patients (5.1%) had an aberrant right subclavian artery behind the trachea, requiring mobilization of the artery in 3 patients and translocation/reimplantation to the carotid artery or ascending aorta in the other 2 patients. 54 patients (55.1%) had an associated tracheal diverticulum that was resected flush with the trachea.

Median days on the ventilator after surgery were 4 days (range 0–81 days). Median total intensive care unit (ICU) stay was 8 days (range 1–134 days). Median total hospital length of stay was 20.5 days (range 3–232 days). There were no significant early complications including hemorrhage or infection.

Median follow-up was 5 months (range 0.25–36 months). There were statistically significant improvements in clinical symptoms postoperatively, including cough, barking cough, noisy breathing, prolonged and recurrent respiratory infections, transient respiratory distress requiring positive pressure, oxygen dependence, blue spells, and ALTEs ($p < 0.001$), as well as ventilator dependence ($p = 0.04$) (Fig. 1).

Table 1
Tracheomalacia scores.

	Location	Preoperative ($n = 98$)	Postoperative ($n = 64$)	p Value
	T1	80 (70–100)	100 (90–100)	<0.001*
	T2	5 (0–30)	100 (70–100)	<0.001*
	T3	5 (0–25)	100 (70–100)	<0.001*
	Right bronchus	100 (80–100)	100 (100–100)	<0.001*
	Left bronchus	90 (50–100)	100 (85–100)	<0.001*
	Trachea (T1–T3)	120 (80–150)	275 (240–300)	<0.001*
	Mainstem bronchi	160 (130–200)	200 (170–200)	<0.001*
	Total	275 (220–330)	450 (420–500)	<0.001*

Preoperative 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). The asterisk means that the p value is statistically significant.

There was a statistical trend toward improved exercise intolerance postoperatively ($p = 0.07$). At latest follow-up, no patients had recurrence of an ALTE.

64 patients (65.3%) underwent postoperative follow-up evaluation with bronchoscopy. Tracheomalacia scores on bronchoscopy improved significantly in all regions of the trachea and bronchi ($p < 0.001$), with the greatest areas of numerical improvement in the middle (T2) and lower (T3) trachea (Table 1).

9 patients (9.2%) had persistent airway intrusion requiring reoperation, using aortopexy in 6 patients, revision posterior tracheopexy in 2 patients, and anterior tracheopexy in 2 patients. 8 of the reoperations were within 9 months following the index surgery. Kaplan–Meier analysis estimates 90% of patients to be free from reoperation at 12 month follow-up (95% confidence interval 83%–97%).

9 patients (9.2%) had tracheostomies preoperatively, all for severe tracheomalacia. Overall, 11 patients (11.2%) had tracheostomies postoperatively, with no significant difference when compared to preoperatively ($p = 0.41$). This included 4 patients who had been transferred from an outside hospital intubated on mechanical ventilation but without tracheostomies preoperatively. 2 of these patients had bilateral vocal cord paralysis requiring tracheostomy. The other 2 patients had associated severe CHD and had tracheostomies placed for prolonged intubation and ventilator weaning. 5 patients with preoperative tracheostomies on the ventilator were able to wean to room air on tracheostomy collar postoperatively. At latest follow-up, 2 patients had undergone tracheostomy decannulation, 1 patient at 2 months and 1 patient at 10 months following additional laryngotracheal reconstruction for subglottic stenosis.

1 patient death occurred 2 months postoperatively in a 5 month old former 34 weeker male with severe CHD, including Raghib complex with a left superior vena cava draining into the left atrium, and ventricular septal defect with pulmonary vein stenosis. His airway was significantly improved on postoperative bronchoscopy with a tracheomalacia score of 460 from 40 preoperatively. However, he developed refractory cardiopulmonary and multisystem organ failure despite maximal oscillatory ventilator settings and inotropic and vasopressor support.

Tracheomalacia scores were negatively correlated with the presence of most clinical symptoms, as in the lower the tracheomalacia score by percentage of open airway, the worse the symptoms. Preoperative total tracheomalacia scores were significantly lower in patients with cough and barking cough (median 225 vs. 345, $p < 0.001$), noisy breathing (median 300 vs. 450, $p < 0.001$), prolonged respiratory infection (median 270 vs. 420, $p < 0.001$), recurrent respiratory infections (median 280 vs. 425, $p < 0.001$), transient respiratory distress requiring positive pressure (median 285 vs. 375, $p < 0.001$), ventilator dependence (median 225 vs. 345, $p = 0.002$), blue spells (median 280 vs. 360, $p < 0.001$), and ALTEs (median 280 vs. 350, $p < 0.001$). Postoperative tracheomalacia scores were significantly lower in patients with exercise intolerance (median 375 vs. 450, $p = 0.05$) and blue spells (median 430 vs. 463, $p = 0.029$). There were no significant differences in preoperative (median 330 vs. 340, $p = 0.46$) scores in patients with exercise intolerance, and preoperative (median 300 vs. 340, $p = 0.99$) or postoperative (median 475 vs. 450, $p = 0.71$) scores in patients with tracheostomies.

3. Discussion

Tracheomalacia is the most common congenital tracheal abnormality [5]. Its reported incidence of 1 in 2100 children is likely an underestimation given that tracheomalacia is often underdiagnosed in the pediatric population [6]. Patients with chronic respiratory symptoms and airflow limitations are often misdiagnosed and incorrectly treated for asthma, reactive airway disease or croup [7]. In addition, tracheomalacia is a common respiratory problem among patients with EA/TEF. Older studies report a prevalence of 11%–33% in this population, likely an underestimation given the wide spectrum of disease and common misdiagnosis, with a recent study reporting tracheomalacia in 87% of EA patients [8–11]. The early and accurate diagnosis of tracheomalacia is important because excessive airway collapse or obstruction leads to ineffective ventilation and clearance of secretions, resulting in frequent respiratory infections, possibly progressing to permanent lung damage in 27% of patients by 8 years of age, and in the most severe cases, blue spells and ALTEs [3–5,11,12]. Many of these children are on repeated courses of antibiotics and steroids that likely affect their quality of life and impair growth.

Currently, there are no definitive standardized guidelines for the evaluation, diagnosis, and treatment of severe tracheomalacia, including grading of symptom severity, criteria for radiographic or endoscopic

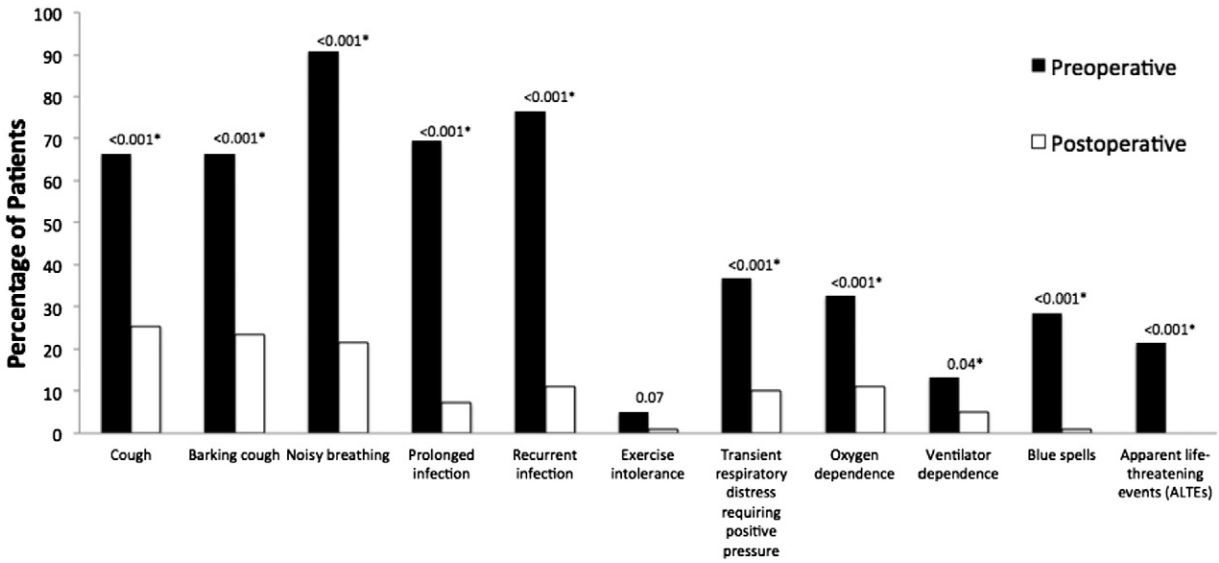


Fig. 1. Preoperative and postoperative clinical symptoms.

evaluation, medical treatment, and surgical approach [1,3,7]. According to the 2012 Cochrane review, there is no evidence to support any one therapy over another for the treatment of tracheomalacia [13].

Our multidisciplinary care team has made efforts to standardize the diagnosis and treatment of tracheomalacia at our institution by creating a standardized reporting and scoring system by anatomic region for endoscopic evaluation [1,4]. 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. A standardized scoring system allows for more effective communication among care providers, as well as for longitudinal airway assessment and its correlation with clinical symptomatology.

Surgical options for the treatment of tracheomalacia include aortopexy (open or thoracoscopic), tracheal resection, external stabilization, and more recently, direct anterior and/or posterior tracheopexy, first reported by our group [1–3,7,14–18]. In a recent meta-analysis, aortopexy was effective in clinically improving more than 80% of children, however 8% showed no improvement, 4% had worsening of their symptoms, and 6% died [16]. Based on endoscopic evaluation, we believe that aortopexy often does not directly address malformed tracheal cartilage and dynamic posterior membranous airway intrusion. In this series, we show that posterior tracheopexy is clinically effective in treating severe tracheomalacia with posterior intrusion with significant improvements in clinical symptoms and degree of airway collapse on bronchoscopy. 18.4% of patients in our series underwent concomitant aortopexy or other pexy procedures. Performing a posterior tracheopexy markedly improves the bronchoscopic success of aortopexy because the posterior wall is fixed to the spine. Most of the patients with persistent airway intrusion requiring reoperation were treated with aortopexy. We favor an individualized approach to optimize each patient's airway and simultaneous or staged procedures as indicated.

There are a number of limitations to this study. Retrospective chart review was used to collect the data, including preoperative and postoperative clinical symptoms and bronchoscopy findings. Although patients are followed 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 three primary operating surgeons. One study in adults showed appropriate interobserver and intraobserver reliability in flexible bronchoscopy, however less is known in the pediatric population [19]. 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 a standardized scoring system to demonstrate resolution of tracheomalacia postoperatively. Tracheomalacia scores tended to correlate with clinical symptoms, and ultimately, the combination of the two is best used together to evaluate the patient. Our study cohort included a heterogeneous group of complex patients requiring adjunct therapies that may have contributed to outcomes and confounded the influence of surgical treatment alone. Follow-up intervals were relatively short-term and variable.

In conclusion, we report the largest series in the pediatric literature on posterior tracheopexy to directly address posterior membranous tracheal intrusion in severe tracheomalacia, and demonstrate significant improvement or resolution of clinical symptoms and airway collapse on bronchoscopy. Further studies are needed to follow long-term outcomes of this technique. Given the heterogeneity and complexity of this patient population, and limited consensus on optimal management strategies to improve patient outcomes, we strongly advocate for a standardized approach to the evaluation of tracheomalacia and individualized patient care in multidisciplinary centers specializing in pediatric airway disorders.

Acknowledgment

H.F.S. was supported by the Joshua Ryan Rappaport Fellowship of the Department of Surgery at Boston Children's Hospital.

Appendix A. Discussions

Hester Shieh, MD

HESTER SHIEH, MD: Good morning. Thank you for the opportunity to present our work. We have no disclosures. Tracheomalacia is characterized by dynamic airway collapse with anterior vascular compression, posterior membranous tracheal intrusion, or both. Severe tracheomalacia refers to dynamic narrowing of the intrathoracic airway during expiration in spontaneously breathing patients. Tracheomalacia is an underestimated disease that is commonly misdiagnosed. It is present in up to 80% of esophageal atresia patients. Excessive airway collapse leads to ineffective ventilation, poor clearance of secretions, and a wide spectrum of respiratory symptoms that can progress to bronchiectasis in a quarter of EA patients by eight years of age. In terms of surgical treatment, aortopexy addresses anterior vascular compression by indirectly elevating the anterior wall of the trachea, but does not directly address posterior membranous tracheal intrusion. We previously reported a small series of patients who underwent direct anterior or posterior tracheopexy for severe tracheomalacia with promising short-term results. In posterior tracheopexy, the posterior tracheal membrane is fixed to the anterior longitudinal spinal ligament under direct bronchoscopic guidance. In this study, we review patients who underwent posterior tracheopexy for severe tracheomalacia with posterior intrusion to determine if there were symptomatic improvements and bronchoscopic evidence of improvement in airway collapse. All patients who underwent posterior tracheopexy from October 2012 to March 2016 were retrospectively reviewed. Clinical symptoms, tracheomalacia scores, and persistent airway intrusion requiring reoperation, were collected. Clinical symptoms included a spectrum of respiratory symptoms, from cough and infections to oxygen and ventilator dependence, to blue spells and apparent life-threatening events. To standardize tracheomalacia anatomy and severity, the tracheobronchial tree was evaluated by anatomic region: trachea broken up into T1, T2, and T3, as well as right and left mainstem bronchi. The percentage of open airway was scored out of 100 at each of the five regions with a maximum score of 500. Ninety-eight patients underwent posterior tracheopexy at median age 15 months. Eighty eight percent were associated with EA. About a third had associated cardiac disease. Sixty nine percent had a prior EA repair and 8% had a prior aortopexy. Upper airway anomalies were common, with laryngeal clefts in 20% of patients. The middle and lower trachea were the most severely affected preoperatively, with median scores of five out of 100 at each region. Generally, patients with associated esophageal disease underwent right posterior thoracotomy, whereas those with cardiac disease underwent sternotomy. All patients underwent posterior tracheopexy with intraoperative bronchoscopic guidance. Fifty-five percent had an associated tracheal diverticulum that was resected flush with the trachea. Eighteen percent of patients underwent concomitant airway procedures, mostly aortopexy, left mainstem bronchopexy, or anterior tracheopexy. Median days on the ventilator were four days, ICU length of stay eight days, and hospital length of stay 20.5 days. There were no significant early complications including hemorrhage or infection. Median follow-up was five months. Anatomically, tracheomalacia scores on bronchoscopy improved significantly in all regions of the trachea and bronchi, with the greatest areas of numerical improvement in the middle and lower trachea. Clinically, there are significant improvements in nearly all symptoms postoperatively,

with a trend toward improved exercise intolerance. At latest follow-up, no patients had recurrence of an ALTE. Nine percent had persistent airway intrusion requiring reoperation, usually with aortopexy. One patient death occurred two months postoperatively in a patient with severe cardiac disease. In conclusion, posterior tracheopexy is effective in treating severe tracheomalacia with significant improvements in clinical symptoms and degree of airway collapse on bronchoscopy. We advocate for a standardized approach to the evaluation and treatment of tracheomalacia in multidisciplinary centers specializing in pediatric airway disorders. Thank you.

DR. CATY: The abstract is now open for questions and I'll start with the first question. The diverticulum that was found in about half of your patients, was that related to prior tracheoesophageal fistula repair?

DR. SHIEH: Yes. Many were related to prior type C repairs.

DR. CATY: And has posterior tracheopexy become your first line of treatment and how do you tailor the type of widening of the trachea that you perform in these patients?

DR. SHIEH: For tracheomalacia with posterior intrusion, it is our preferred treatment and using intraoperative bronchoscopy is key in terms of making sure that the airway remains open.

DR. LANGHAM: Max Langham, Memphis. Dr. Shieh, this is very cool stuff and I had two questions. Your follow-up probably was from a range of times, but do you have how long the follow-up was on some of your patients?

DR. SHIEH: Yes, it ranged from less than a month to 36 months.

DR. LANGHAM: Okay. And is there any change in the symptoms in the folks that have been out three years versus those that are out six months?

DR. SHIEH: The symptoms reported were at latest follow-up, but it seemed that people did well and same with the group who had longer follow-up as well.

DR. LANGHAM: The second question is the procedure requires mobilization and movement of the esophagus and in your complications, there is nothing about dysphagia, problems with recurrent laryngeal nerve or other things that are not directly related to the aortopexy or the tracheopexy, so I was curious as to whether or not you had any esophageal symptoms in any of the patients.

DR. SHIEH: I think that's something we are looking at further, postop dysphagia.

References

[1] Bairdain S, Smithers CJ, Hamilton TE, et al. Direct tracheobronchopexy to correct airway collapse due to severe tracheobronchomalacia: short-term outcomes in a series of 20 patients. J Pediatr Surg 2015;50:972–7.

[2] Jennings RW, Hamilton TE, Smithers CJ, et al. Surgical approaches to aortopexy for severe tracheomalacia. J Pediatr Surg 2014;49:66–70 [discussion 70-1].

[3] Fraga JC, Jennings RW, Kim PC. Pediatric tracheomalacia. Semin Pediatr Surg 2016; 25:156–64.

[4] Ngerncham M, Lee EY, Zurakowski D, et al. Tracheobronchomalacia in pediatric patients with esophageal atresia: comparison of diagnostic laryngoscopy/bronchoscopy and dynamic airway multidetector computed tomography. J Pediatr Surg 2015;50: 402–7.

[5] Boogaard R, Huijsmans SH, Pijnenburg MW, et al. Tracheomalacia and bronchomalacia in children: incidence and patient characteristics. Chest 2005;128: 3391–7.

[6] Fischer AJ, Singh SB, Adam RJ, et al. Tracheomalacia is associated with lower FEV1 and *Pseudomonas* acquisition in children with CF. Pediatr Pulmonol 2014;49: 960–70.

[7] Bairdain S, Zurakowski D, Baird CW, et al. Surgical treatment of trachebronchomalacia: a novel approach. Paediatr Respir Rev 2016;19:16–20.

[8] Spitz L, Kiely E, Brereton RJ. Esophageal atresia: five year experience with 148 cases. J Pediatr Surg 1987;22:103–8.

[9] Slany E, Holzki J, Holschneider AM, et al. Tracheal instability in tracheo-esophageal abnormalities. Z Kinderchir 1990;45:78–85.

[10] Filler RM, Messineo A, Vinograd I. Severe tracheomalacia associated with esophageal atresia: results of surgical treatment. J Pediatr Surg 1992;27:1136–40 [discussion 1140-1].

[11] Cartabuke RH, Lopez R, Thota PN. Long-term esophageal and respiratory outcomes in children with esophageal atresia and tracheoesophageal fistula. Gastroenterol Rep 2016;4:310–4.

[12] Carden KA, Boiselle PM, Waltz DA, et al. Tracheomalacia and tracheobronchomalacia in children and adults: an in-depth review. Chest 2005;127:984–1005.

[13] Goyal V, Masters IB, Chang AB. Interventions for primary (intrinsic) tracheomalacia in children. Cochrane Database Syst Rev 2012;10:CD005304.

[14] van der Zee DC, Straver M. Thoracoscopic aortopexy for tracheomalacia. World J Surg 2015;39:158–64.

[15] Perger L, Kim HB, Jaksic T, et al. Thoracoscopic aortopexy for treatment of tracheomalacia in infants and children. J Laparoendosc Adv Surg Tech A 2009; 19(Suppl. 1):S249–54.

[16] Torre M, Carlucci M, Speggiorin S, et al. Aortopexy for the treatment of tracheomalacia in children: review of the literature. Ital J Pediatr 2012;38:62.

[17] Weber TR, Keller MS, Fiore A. Aortic suspension (aortopexy) for severe tracheomalacia in infants and children. Am J Surg 2002;184:573–7 [discussion 577].

[18] Dave S, Currie BG. The role of aortopexy in severe tracheomalacia. J Pediatr Surg 2006;41:533–7.

[19] Majid A, Gaurav K, Sanchez JM, et al. Evaluation of tracheobronchomalacia by dynamic flexible bronchoscopy. A pilot study. Ann Am Thorac Soc 2014;11:951–5.