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Categorization and repair of recurrent and acquired tracheoesophageal fistulae occurring after esophageal atresia repair☆

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ABSTRACT

Purpose: Recurrent trachea-esophageal fistula (recTEF) is a frequent (5%–10%) complication of congenital TEF (conTEF) and esophageal atresia (EA) repair. In addition, postoperative acquired TEF (acqTEF) can occur in addition to or even in the absence of prior conTEF in the setting of esophageal anastomotic complications. Reliable repair often proves difficult by endoluminal or standard surgical techniques. We present the results of an approach that reliably identifies the TEF and facilitates airway closure as well as repair of associated tracheal and esophageal problems.

Methods: Retrospective review of 66 consecutive patients 2009–2016 (55 referrals and 11 local) who underwent repair via reoperative thoracotomy or cervicotomy for recTEF and acqTEF (IRB P00004344). Our surgical approach used complete separation of the airway and esophagus, which reliably revealed the TEF (without need for cannulation) and freed the tissues for primary closure of the trachea and frequently resection of the tracheal diverticulum. For associated esophageal strictures, stricturoplasty or resection was performed. Separation of the suture lines by rotational pexy of the both esophagus and the trachea, and/or tissue interposition were used to further inhibit re-recurrence. For associated severe tracheomalacia, posterior tracheopexy to the anterior spinal ligament was utilized.

Results: The TEFs were recurrent (77%), acquired from esophageal leaks (26%), in addition to persistent or missed H-type (6%). Seven patients in this series had multiple TEFs of more than one category. Of the acqTEF cases, 6 were esophagobronchial, 10 esophagopulmonic, 2 esophagotracheal (initial pure EA cases), and 2 from a gastric conduit to the trachea. Upon referral, 18 patients had failed endoluminal treatments; and open operations for recTEF had failed in 18 patients. Significant pulmonary symptoms were present in all. During repairs, 58% were found to have a large tracheal diverticulum, and 51% had posterior tracheopexy for significant tracheomalacia. For larger esophageal defects, 32% were treated by stricturoplasty and 37% by segmental resection. Rotational pexy of the trachea and/or esophagus was utilized in 62% of cases to achieve optimal suture line separation. Review with a mean follow-up of 35 months identified no recurrences, and resolution of pulmonary symptoms in all. Stricture treatment required postoperative dilations in 30, and esophageal replacement in 6 for long strictures. There was one death.

Conclusion: This retrospective review of 66 patients with postoperative recurrent and acquired TEF following esophageal atresia repair is the largest such series to date and provides a new categorization for postoperative TEF that helps clarify the diagnostic and therapeutic challenges for management.

Following repair of esophageal atresia (EA) with a tracheoesophageal fistula (TEF) the communication recurs in up to 5%–10% of cases. [1–9] Repair of recurrent tracheoesophageal fistula (recTEF) poses several problems for the surgeon including accomplishing a safe and effective repair in a reoperative setting, a yet higher risk of re-recurrence, and, in many cases, the treatment of associated complex problems of esophageal stricture, anastomotic defects and airway lesions. Another complicating factor presents with cases that have a TEF that is difficult to localize and/or in a different location than the original TEF.

We have developed a new classification system for postoperative "recurrent" TEFs that more accurately reflect their etiology and anatomy. Congenital TEFs (conTEFs) are those which persist after repair because they were either missed (such as a proximal TEF which was missed when a distal TEF was repaired), or the repair attempt was incomplete leaving the conTEF intact. These are present immediately after the repair attempt. The second category is the recurrent TEF

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(recTEF) that occurs in the same location as the TEF repaired at the primary operation. These most commonly follow Type C esophageal atresia repair with the TEF forming between the esophageal anastomosis and the tracheal diverticulum, but can also be seen after proximal H-type TEF repair. The third category is the acquired TEF (acqTEF) which forms along a new pathway, with a new location on either the airway side or the digestive side, or both. These include communications between the esophageal anastomosis and the pulmonary parenchyma, a segmental bronchus, or the trachea. These also include communications between a colon or gastric conduit and the respiratory system, anywhere from the trachea to bronchi to pulmonary parenchyma. (Fig. 1) Each of these can have different challenges in evaluation and management.

The purpose of this study was to review postoperative conTEF, recTEF and acqTEF patients and report on the preoperative characteristics of these patients and our method of evaluation and surgical repair, and the outcomes of this complex group of patients. The patient review allowed an assessment of the effectiveness of our approach for postoperative TEF repair as well as our results treating both the fistulae and complex associated lesions. This article reviews the largest single institution series for postoperative TEF patients to date, and additionally describes our approach and some of the techniques we used for preventing re-recurrence including rotation esophagoplasty and posterior tracheopexy.

1. Methods

Sixty-six consecutive patients with postoperative TEF referred to the Esophageal and Airway Treatment (EAT) Center and repaired at Boston Children's Hospital (2009–2016) were reviewed. Patient data collected included: original type of EA/TEF lesion with method of repair, and initial complications of esophageal leak and/or stricture. The postoperative TEF data included the apparent time of occurrence, related symptoms, means of identification, prior localization techniques, and prior attempted endoscopic and operative recTEF repairs. At our EAT Center, the components of operative repair, surgical results, length of follow-up and patient outcome were reviewed (IRB Protocol P00004344). Four surgeons from the EAT Center (JF, RJ, TH, and JS) comprised the operative team for these cases, generally working with two attending surgeons at a time.

Our evaluation for all patients suspected to have a postoperative TEF included an esophagram and endoscopy – comprised of rigid and flexible bronchoscopy and esophagoscopy – to determine the anatomic location of the fistula. (Fig. 2) Other tracheal findings were assessed including the size of the residual diverticulum from the original repair site, and detailed description of tracheomalacia by static and dynamic bronchoscopy. Bronchoscopy with spontaneous breathing is absolutely critical as tracheomalacia is easily underestimated by static bronchoscopy in a deeply anesthetized patient. A CT scan of the chest including dynamic 3D and 4D reconstructions of the airway was used to examine and classify tracheomalacia, to identify the anatomic relationships of the trachea and esophagus to the major mediastinal vasculature, and to identify vascular anomalies including aberrant right subclavian arteries and vascular rings. CT scans were selectively used for more complex cases that had significant tracheomalacia by initial bronchoscopy, suspected vascular anomalies based on prior echocardiograms, or numerous prior thoracotomies. (Fig. 3).

Repair was by an open surgical approach, either thoracotomy or cervicotomy based on the above evaluation, with complete mobilization of the lung and then meticulous sharp dissection of the airway and esophagus, avoiding ischemic injury to the esophagus and trachea. We did not generally utilize techniques of catheter or wire localization of the fistula. In fact, in cases of acquired fistulae to the bronchi and lung parenchyma, or cases of multiple fistulae, this was not feasible. Complete dissection of the esophagus reliably revealed the airway end of the fistula by the air leak with ventilation. Flexible bronchoscopy was used during the repair of the trachea to confirm fistula closure and that a flush resection and repair of any residual tracheal diverticulum was accomplished. Diverticulum resection occasionally required a flap closure of the membranous trachea to be repaired if the luminal orifice of the diverticulum was very large. This operative method revealed the various postoperative acquired TEFs to the pulmonary parenchyma and distal airways as well, and freed the tissues for a well-visualized and tension-free closure of both sides of the fistula. Posterior tracheopexy to the anterior spinal ligament was used to help cover the tracheal repair and separate it from the esophageal repair. This also has the advantage of correcting tracheomalacia by preventing dynamic posterior intrusion of the posterior tracheal membrane into the tracheal lumen by fixing the membranous trachea to the prevertebral fascia. [10] (Fig. 4).

For the resultant esophageal defects, transverse orientation of the esophageal repair was preferred to minimize esophageal stenosis. For significant associated esophageal strictures, stricturoplasty or stricture resection was performed. In six cases, longer esophageal strictures prevented primary esophageal repair. These were staged using the Foker process for traction induced esophageal growth in 3 cases, and

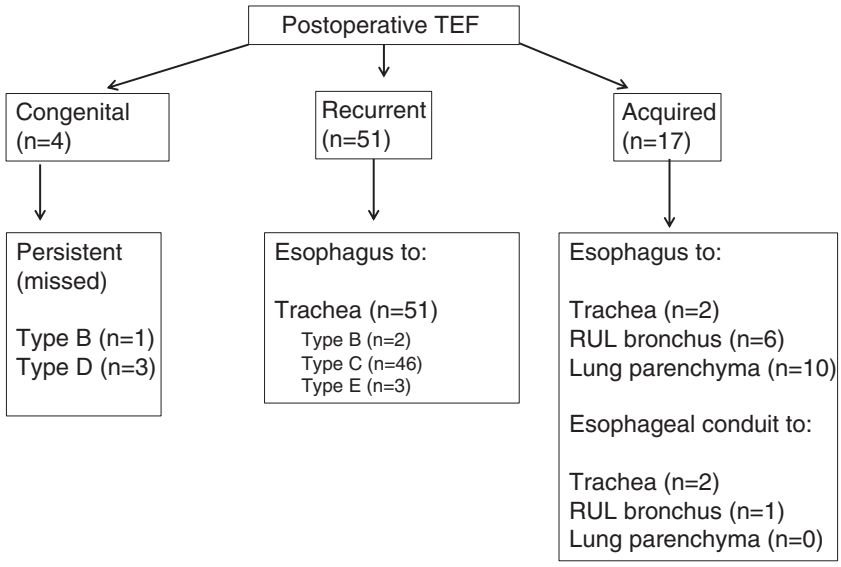

Fig. 1. Categorization of tracheoesophageal fistulae (TEF). [Types B, C, D, and E refer to Gross classification of esophageal atresia types].