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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

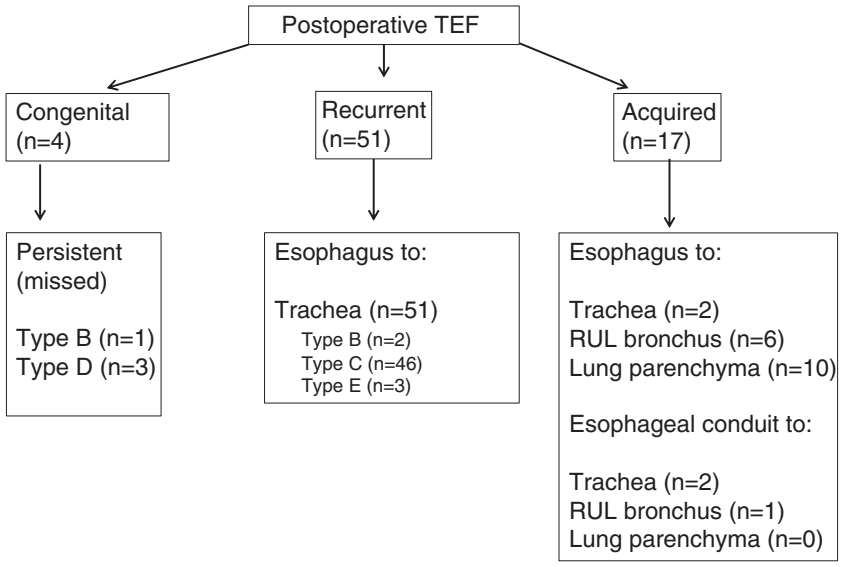

Fig. 1. Categorization of tracheoesophageal fistulae (TEF). [Types B, C, D, and E refer to Gross classification of esophageal atresia types].

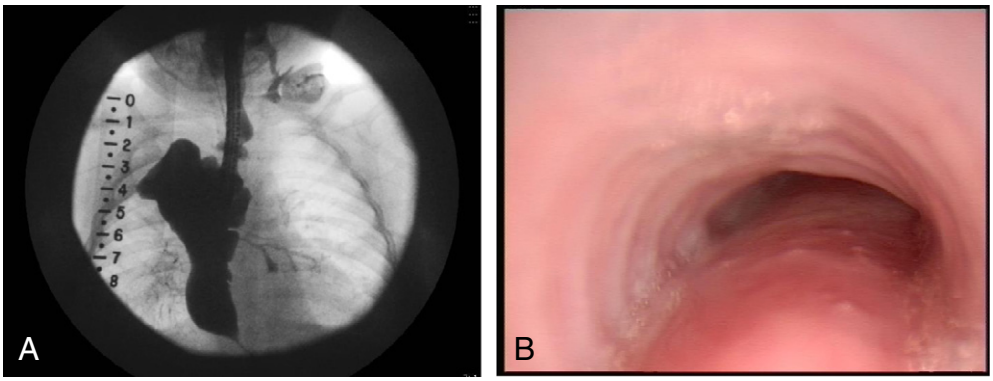

Fig. 2. Studies for patient with recTEF, original type C. A. Complex chronic esophageal leak cavity in addition to recTEF. B. Bronchoscopy showing severe tracheomalacia from posterior intrusion of the membranous trachea.

cervical esophagostomy in 4 cases with subsequent jejunal interposition. [11,12].

To further inhibit TEF re-recurrence, rotation of the esophagus and trachea was used to increase suture line separation. This was accomplished by posterior tracheopexy as described above for the tracheal side of the fistula. This type of slight rotation of the tracheal closure is designed to prevent recurrent fistulization, even in the setting of a recurrent esophageal leak. For simple closure of smaller esophageal defects following TEF division, similar rotational pexy of esophageal closure away from the trachea serves the same purpose. Tissue interposition was added for some cases if the tracheal and esophageal suture lines remained closely apposed. These were generally small local flaps of pleural and scar tissue or sometimes local lymph nodes. A partial sternocleidomastoid muscle flap was used for one case of recurrent cervical H-type fistula. We did not perform any more elaborate forms of tissue interposition such as intercostal or pericardial flaps.

2. Results

From 2009 to 2016, sixty-six consecutive patients were treated for postoperative conTEF, recTEF and acqTEF in the setting of prior repairs for esophageal atresia. Fifty-five patients were referred from other institutions and eleven were from our hospital. Of the 11 patients from our own hospital, 8 had acqTEF in the setting of prior repairs for long gap or complicated esophageal atresia, one had recurrent cervical H-type TEF, one had type C EA/TEF with initial ligation of the TEF in continuity for an unstable premature neonate, and one had recTEF following thoracoscopic repair of type C EA/TEF. See Table 1 for patient demographics and preoperative characteristics. Prematurity was present in 62% of cases, and low birth weight in 55%. While the majority of the original esophageal atresia cases were type-C (79%), all types were represented. A large portion of patients were found to have had either an early postoperative esophageal leak (48%) and/or stricture (77%) following the primary repair, and had esophageal dilations (59%) before diagnosis of the postoperative TEF. The time to identification of the postoperative TEFs following the index or subsequent repair operations was quite variable, ranging from within 5 days to over a decade. The most common presenting symptoms were respiratory in nature including coughing, choking, increased pulmonary secretions, and recurrent pneumonia. The patient age for repair at our institution ranged from 3 weeks to 18 years (mean 31.5 months, median 16 months). (See Table 1).

Fig. 3. CT scan with 3D reconstruction for patient with moderate to severe tracheomalacia. This patient had a missed proximal conTEF after prior type C EA/TEF repair.

Twenty-five patients (38%) had prior open surgical and/or endoluminal procedures for recTEF before referral to our center. The number of recTEF for each patient ranged from 1 to 12, with a mean of 2.3. However, distinguishing the number recurrences of the TEF for each patient rather than actual persistence of the TEF in the setting of numerous attempts at prior treatment (especially endoluminal) is probably not realistic. Of the patients with prior operative attempts to eradicate the recTEF, eight patients had two or more redo operations each before referral. Included in this group is one patient who underwent slide tracheoplasty via cervical approach but continued to have an esophagopulmonic fistula (acqTEF) from the esophageal anastomosis to the right upper lobe (Fig. 3), and another who had four prior redo operations including a colon-esophageal patch and right upper lobectomy, but continued to have a fistula from the esophageal anastomosis to the right upper lobe bronchus (acqTEF) (Fig. 4). Two patients with prior gastric pull-ups developed numerous recTEF and acqTEF along the back wall of the trachea and right bronchus. Many of these patients who presented with recTEF despite prior operative treatment for the same, did have tissue interpositions as a component of their unsuccessful reoperative strategy, including muscle and pleural flaps, and one case of Surgisis (®) mesh. The prior endoluminal methods utilized included mucosal injury by laser, sclerosis, or cautery, the use of various tissue sealants such as fibrin glue and histoacryl, and one case of placement of an endovascular plug device (See Table 2).

In four cases, patients were found to have a proximal H-type TEF that had initially been missed (conTEF), although one patient had undergone endoscopic treatments at the original more distal type-C TEF site prior to referral to our center. In 17 patients (26%), the postoperative TEF did not occur at the original conTEF site, which we now call acqTEFs. Seven patients were found to have multiple TEFs that fit into more than one category. (See Table 3)

Our operative approach is described in detail above, and the technical components itemized in Table 4. Airway closures involved the membranous trachea for 56 patients (86%), right bronchus for 9%, right upper

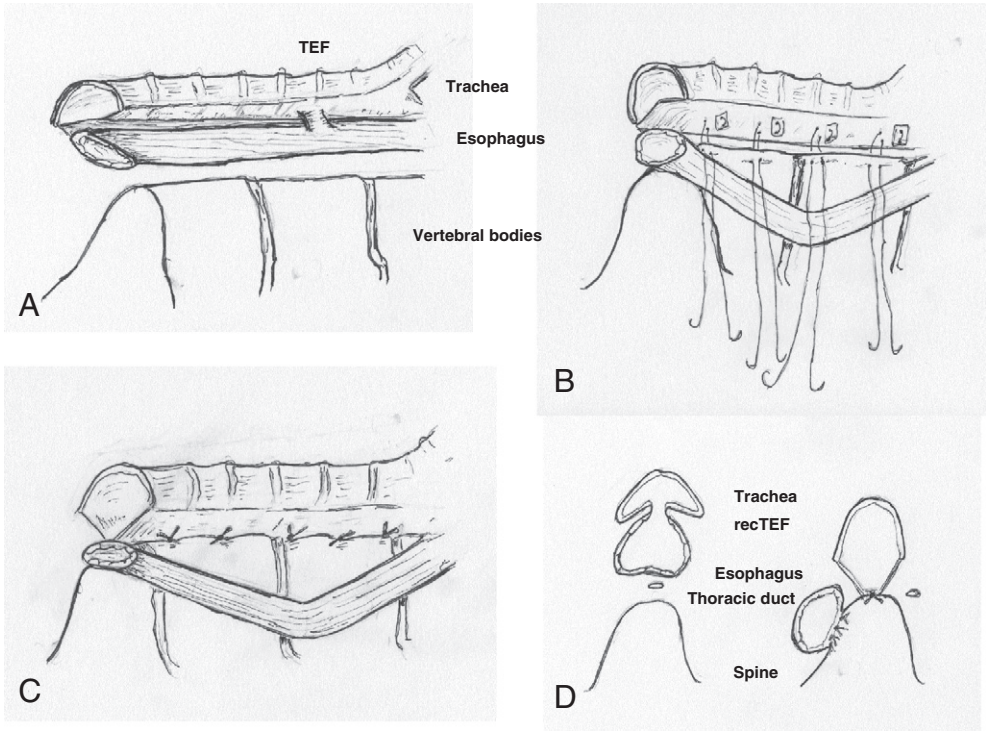

Fig. 4. Posterior tracheopexy. A. Illustrates anatomic relationship of trachea, esophagus and spine with recTEF prior to operation. B. RecTEF and tracheal diverticulum resected, esophagus retracted to the right, posterior tracheopexy sutures placed in midline of membranous trachea to anterior spinal ligament. C. Posterior tracheopexy sutures tied and membranous trachea now fixed to anterior spinal ligament, enlarging the tracheal lumen and preventing dynamic posterior intrusion of the membranous trachea. D. Cross-sectional view of same procedure, including rotation esophagoplasty. Drawings courtesy of Dr. Neil Feins.

Table 1
Patient demographics.

Preoperative characteristics	Number of patients (%, N = 66)
M/F	36/30
Gestational age	
<32 weeks	6 (9%)
32–35 weeks	20 (30%)
36–38 weeks	15 (23%)
>38 weeks	25 (38%)
Birthweight	
<1.5 kg	5 (8%)
1.5–2.0 kg	12 (18%)
2.1–2.5 kg	19 (29%)
2.6–3.0 kg	18 (27%)
>3 kg	12 (18%)
Esophageal atresia type	
Type A long gap	7 (11%)
Type B long gap	3 (5%)
Type C	49 (74%)
Type C long gap	3 (5%)
Type D	1 (2%)
Type E (H-type)	3 (5%)
Original repair approach	
Thoracotomy	57 (86%)
Cervicotomy	3 (5%)
Thoracoscopy	6 (9%)
LGEA subgroup	
Foker process	10 (15%)
Primary repair	1 (2%)
Gastric tube	1 (2%)
Kimura procedure	1 (2%)
Complications of original repair	
Anastomotic leak	32 (48%)
Anastomotic stricture	51 (77%)
Treated by dilations	39 (59%)
Treated by stent	7 (11%)
Age of repair for recTEF at our center	3 weeks–18 y (mean 31.5 mo; median 16 mo)

lobe lung wedge resection for one patient, and direct suture repair for lung parenchymal involvement in 15%. In 38 patients (58%) the tracheal diverticuli were resected flush with the tracheal wall. For 33 patients (51%), a posterior tracheopexy was performed in conjunction with the airway repair. (See Table 4)

Esophageal closure techniques are also shown in Table 4. Eighteen patients (28%) had no stricture and therefore simple suture closure of the esophageal side of the TEF. The strategy for esophageal stricture management was directed by length of the stricture, using simple stricturoplasty in 21 cases (32%) and segmental stricture resection in 24 cases (37%). Three patients with initial type-C esophageal atresia had developed long gap esophageal defects from severe strictures, and

Table 2
Attempted procedures for recTEF prior to referral.

Characteristics of prior recTEF repairs	Number of patients (%, N = 66)
Approach	
Open thoracotomy	15 (23%)
Tissue interposition	7 (11%)
Surgisis	2 (3%)
Muscle flap	4 (6%)
Pleural flap	2 (3%)
Cervicotomy	3 (5%)
Slide tracheoplasty	1 (2%)
Tissue interposition	1 (2%)
Endoscopic	18 (27%)
Laser	3 (5%)
Cauterization	2 (3%)
Fibrin glue	7 (11%)
Tisseal	2 (3%)
Collagen	1 (2%)
Surgisis	4 (6%)
Histoacryl	1 (2%)
Sclerosis	2 (3%)
Endovascular plug device	1 (2%)
Number of recTEFs per patient	Range: 1–12, Mean: 2.3

Table 3
Categorization of postoperative acquired and recurrent TEF.

Postoperative TEF	Number of Patients (%, N = 66)
Recurrence to initial TEF site (recTEF)	51 (77%)
Original type-C location	46 (70%)
Including long tracheal diverticulum	37 (56%)
Original H-type location	3 (5%)
Original proximal pouch TEF for type-B	2 (3%)
From gastric conduit to trachea	1 (2%)
Missed proximal H-type TEF (conTEF)	4 (6%)
Acquired TEF to new airway or digestive location (acqTEF)	17 (26%)
Esophagus to membranous trachea	2 (3%)
Esophagus to RUL bronchus	6 (9%)
Esophagus to lung parenchyma	10 (15%)
1 case from esophageal stent erosion	1 (2%)
Gastric conduit to trachea	2 (3%)
Gastric conduit to RUL bronchus	1 (2%)

(Note: Seven patients had multiple recTEFs within multiple categories).

Table 5
Post-operative outcomes.

Outcomes	Number of patients (%, N = 66)
Follow-up 3–87 months (median 29.5)	
Re-recurrence of TEF	0%
Resolution of pulmonary symptoms	65 (98%)
This includes perioperative death for total count, all other patients had resolution of symptoms	
Esophageal anastomotic leak	6 (9%)
(5 with spontaneous resolution, 1 required reoperation)	
Need for esophageal dilations	30 (45%)
Number of dilations each: range 1–15, mean 4.7	
Cervical esophagostomy	4 (6%)
Two still awaiting jejunal interposition	
Esophageal replacement by jejunal interposition	4 (6%)
Death	1 (2%)
Aortic injury in patient with right aortic arch	

were treated by the Foker process using traction induced growth. Two of these cases later required esophagectomy, because of a refractory stricture in one and chronic esophageal leak for the second. They have both undergone jejunal interposition. (See Table 5) The third more recent case had successful primary esophageal repair by the Foker process with the anastomosis transposed to the thoracic inlet – away from the lung parenchyma of the right upper lobe, the site of 4 prior TEF recurrences. A total of four patients (including one of the above mentioned cases initially repaired using Foker process) required cervical esophagostomy and were subsequently reconstructed by jejunal interposition.

The interval of follow-up ranged from 3 to 87 months, with a median of 29.5 months. No patient has been found to have a recurrence of the TEFs we repaired, and all patients had resolution of the pulmonary symptoms. Six patients (9%) had an early contained esophageal anastomotic leak; 30 patients (45%) have required esophageal dilations in the postoperative period. Six patients have or will require esophageal replacement, as described above. Our group includes one death from operative bleeding related to an aortic injury. This patient had a right-sided aortic arch, and the esophageal stricture and recTEF were adherent to the aorta (the initial operation had been by right thoracotomy, as was our reoperation). Although that patient survived the immediate operation via aortic patch repair on cardiopulmonary bypass, they suffered irreversible neurologic injury. (Table 5)

Table 4
Technical components of repair.

Operative technique	Number of patients (%, N = 65)
Airway	
Primary tracheal closure	56 (86%)
Including resection of tracheal diverticulum	38 (58%)
Right bronchus repair	6 (9%)
Lung wedge resection	1 (2%)
Suture repair of lung parenchymal air leaks	10 (15%)
Rotation of membranous trachea toward spine	40 (62%)
Including posterior tracheopexy for tracheomalacia	33 (51%)
Esophagus	
Transverse oriented closure of small defect	18 (28%)
Stricturoplasty by transverse closure	21 (32%)
Long linear closure	1 (2%)
Segmental resection	24 (37%)
Foker process for growth	3 (5%)
Cervical esophagostomy	4 (6%)
Rotation of esophageal closure away from trachea	31 (48%)
Tissue interposition	24 (37%)
Muscle flap for cervical H-type	1 (2%)
Intercostal muscle flap (preserved from prior operation)	1 (2%)
Small pleural flap	13 (20%)
Azygous vein	1 (2%)
Local lymph nodal tissue	9 (14%)
Local scar tissue	5 (8%)
Thymus glandular tissue	1 (2%)
Surgisis patch	1 (2%)

(Note: N = 65 excludes one patient death for whom recTEF repair was not completed.)
(Also, 7 pts. had TEFs to multiple locations).

3. Discussion

A fistula can occur after initial operative repair of esophageal atresia because of a variety of causes – primarily esophageal anastomotic leak, but also surgical tissue injury, foreign body erosion from sutures or clips, trauma from esophageal dilations, and infection. Once present, the differential pressures between the airway and esophagus facilitate transfer of the contents from one to the other. [13] While the transfer of tracheal contents to the esophagus is probably inconsequential, the transfer of esophageal contents into the airways can have severe consequences. These include chronic cough, choking, cyanosis with feeding, tracheitis, and recurrent pneumonias. Recurrent pneumonias can lead to bronchiectasis, and tracheitis can contribute to progression of tracheomalacia. [14,15] Additionally, recurrent tracheoesophageal fistulae in the context of esophageal atresia are often part of more complex issues including tracheomalacia that impairs airway clearance, tracheal diverticuli that promote pooling of secretions and infection, esophageal strictures that increase the pressure differential between the esophagus and trachea, and gastroesophageal reflux that increases the acidity and bile content of the fluid transferred from the esophagus to the airway producing increased airway irritation. [16] As such, postoperative TEFs must be considered as one part of a complex system and the repair must take the systematic issues into account in order to optimize outcome and success [4].

Regarding mechanisms of postoperative TEF development and categorization, this series has shown several interesting features. Many patients acquired fistulae to a new airway site other than the original congenital TEF location, or indeed in the absence of a congenital TEF. In fact, perhaps it is more accurate to use the terms postoperative esophagobronchial fistula and esophagopulmonic fistula for some of these more complex cases. The commonality found in these cases was that the esophageal anastomosis was involved in the fistulization process in the form of a chronic leak site with or without a coexistent stricture. Whether by stent erosion or ongoing esophageal injury from dilations, when esophageal anastomotic problems persist, they can result in fistulae to other portions of the airway or lung. This clearly illustrates the importance of addressing any ongoing esophageal strictures simultaneous to airway procedures for postoperative TEF, whether endoluminal or surgical. The case in our series of an acqTEF following slide tracheoplasty demonstrates this most pointedly. Despite a very effective tracheal repair strategy, the presence of ongoing esophageal anastomotic issues resulted in a persistent esophagopulmonic fistula

in that case. Additionally, for patients with a suspected postoperative TEF that is elusive to conclusively diagnose and locate, consideration for an acqTEF should be made.

Postoperative conTEFs, recTEFs and acqTEFs present challenges on several fronts. First, diagnosis and localization to prove that a TEF has occurred after a repair can be difficult. [5,6,17–20] Several methods have been described to facilitate diagnosis, essentially based on contrast studies and endoscopy. [13] Within our own series, several patients required multiple rounds of evaluation before the diagnosis was definitively established. The pathogenesis and anatomy of these fistulae imply that they can be intermittent, and therefore more difficult to diagnose at a single investigation. The multiplicity of symptoms with which esophageal atresia patients can present further complicates the diagnosis. Problems of coughing, choking, increased secretions are frequent in these patients, also related to esophageal strictures, dysmotility, GERD, and tracheomalacia. In our series, the comprehensive evaluation described provided a thorough assessment of not only the postoperative TEF (conTEF, recTEF and/or acqTEF), but of related tracheal and esophageal problems to be addressed during the operative intervention.

The operation for either a recTEF or an acqTEF will be more difficult than for a conTEF. Postoperative adhesions and scarring make the operation more difficult and hazardous and tissues less supple. The failure rate, therefore, would be expected to be greater than for conTEF repairs, and multiple reports have born this out. [7] In their 2014 systematic review of management for recTEF, Aworanti and Awadalla very nicely summarized the results from 44 papers on this topic including 165 total patients. The collective refistulization rate was 21% for redo open surgery and 63% for endoluminal techniques. The overall reported mortality was 3.7% for open operations and 1.7% for endoluminal approaches. Esophageal replacement was required in 2.4% of total cases. Of the most recent published reports, refistulization rates still range 8%–17%, with up to 10% mortality. [5–9] And of course, as difficult as these reoperations can be, further subsequent operations for re-recurrences will, predictably, be even more difficult. [7,21].

The above mentioned risks do make endoluminal methods of closure appear as attractive options. Unfortunately, the success rate of endoscopic methods for repair of postoperative TEFs is poor. [7,22] The anatomy of the postoperative TEF likely plays a role here; these are often very short communications, within a system that is constantly subjected to high pressure changes, often with sutures or other foreign bodies present. In the frequent situation of a simultaneous esophageal stricture, this high pressure system is even more pronounced. The wide luminal diameter of the tracheal diverticulum that is often associated with the original type-C TEF and subsequent recTEF also contributes to the low likelihood of successful endoluminal closure. Nonetheless, for the patient that is found to have a very small caliber recTEF that has a longer tract before entering the esophagus, endoluminal methods may still play some role. [22] Certainly, even for these patients however, simultaneous endoscopic dilations of an esophageal stricture may thwart any chance of success by ongoing local trauma and esophageal anastomotic disruption. Lastly, regarding endoluminal treatment, several patients within our series had near fatal airway occlusion episodes following dislodgement of endoscopically injected tissue sealants in the airway. In our series and currently within our practice, we have not been in favor of endoluminal methods of TEF closure, although we do think that this strategy may still have some benefit for certain cases, as described above. We continue to be impressed however that the vast majority of patients with a postoperative TEF have additional issues including tracheal diverticuli, tracheomalacia, and esophageal strictures, all of which together strongly favor an operation that addresses everything at once.

Regarding the conduct of the operation for postoperative TEF, this series illustrates several points. It goes without saying that meticulous dissection is necessary in redo esophageal surgery. Starting the esophageal mobilization in the less scarred distal esophagus and carefully proceeding with sharp dissection for the remainder of the dissection is critical. Endoscopy of both the esophagus and airway can be beneficial during this dissection. Focus on finding and staying just on the esophageal wall prevents injury to the many important structures at risk including the airway, vagus and recurrent laryngeal nerves, thoracic duct, and any aberrant arteries. Complete dissection of the esophagus provides clear localization of the fistula, which is particularly helpful in cases of multiple and/or acquired fistulae. This obviates dependence on precise endoscopic localization or catheterization of the fistula. It should be noted that our evaluation before surgery did provide a very discrete concept of the TEF location(s) for all patients in the study, and informed our choice of operative approach, whether cervicotomy or thoracotomy.

We have found several components useful in effective airway closure following division of the postoperative TEF. Fine absorbable monofilament suture decreases the risk of foreign body reaction. Intraoperative flexible bronchoscopy helps plan and guide airway closure and resection of the tracheal diverticulum, and aids in the prevention of airway stenosis. Bronchoscopy also is critical during the techniques of tracheal rotation and/or posterior tracheopexy, in order to guide suture placement and prevent angulation of the trachea by the pexy procedure. We have preferred this strategy of suture line rotation with posterior tracheopexy over formal tissue interposition in the prevention of re-recurrence of the TEF, particularly since tissue interposition had failed to prevent re-recurrence of the recTEF procedures that had been performed prior to referral to our EAT center. The posterior tracheopexy creates apposition of the tracheal fistula closure suture line to the anterior spinal ligament as an effective strategy to separate this closure from any possible future esophageal anastomotic problems. The posterior tracheopexy procedure is described in detail in our paper regarding tracheomalacia management. [10] The technique actually developed as a consequence and modification of tracheal suture line rotation that originally was not designed to specifically address tracheomalacia, but only rather to help prevent re-recurrence for recTEF patients. We became very impressed however in our subsequent evaluations and long-term follow-up that the technique, when precisely applied, is very effective for eliminating posterior intrusion type tracheomalacia.

In addition to the airway, the esophagus can also be rotated by suture pexy to the thoracic wall or vertebral bodies to prevent direct apposition of the suture lines. Care must be taken to avoid injury to the thoracic duct during these pexy maneuvers. Completion flexible esophagoscopy during the operation is important to make sure the esophagus has not been kinked or excessively twisted. We have used small flaps of local tissue (pleura, scar, and lymph nodes) to cover and further separate the tracheal and esophageal closures, although admittedly this tissue is often of questionable integrity; hence, we feel the rotation maneuvers are of higher importance. Although more formal tissue interpositions can be performed such as pericardium, intercostal muscle, and cervical muscles in the case of cervical H-type fistulae, there are potential disadvantages to all of these approaches such as cardiac herniation, pericarditis, and posterior tracheal compression in the case of larger muscle flaps. [23,24] Muscle and tissue flaps around the esophagus may also contribute to the development of difficult esophageal strictures or extrinsic compression stenosis. Some of the most difficult patients within this series involved cases where prior tissue interposition flaps (primarily intercostal muscle) had not only failed to prevent repeat recurrence of a TEF, but also contributed to severe esophageal strictures.

Regarding the outcomes for patients in this series, we can report that no patient has been found to have a re-recurrence of the TEF with a mean follow-up time of 35 months. From our experience in this patient series and other reports, the most common time to TEF recurrence is within 2–3 months, although much longer intervals are possible. [4–8] We maintain close long-term follow-up with all of our patients treated within the EAT center, which includes surveillance endoscopy and contrast study evaluations when indicated. So, while we cannot exclude the

possibility that some of the patients reported in this study may yet develop a future repeat recTEF or acqTEF, we have not seen this to date. Our patients did have a small rate of esophageal leak (9%) and common need for further esophageal dilations following the TEF repair (45%). We routinely plan for a series of esophageal dilations starting 3 weeks after surgery for many of our patients, especially when a complete esophageal resection and anastomosis were needed. Regarding the need for esophageal replacement in six patients, this highlights the severity of esophageal anastomotic complications that can co-exist in the context of recTEF and acqTEF. Several of the more complicated cases in this study also highlight the possibility of postoperative TEF development following esophageal replacement. For a variety of reasons addressed in other studies, we prefer jejunal interposition as the most ideal esophageal replacement conduit. [25] When positioned substernally to reach the cervical esophagus, issues of future postoperative TEF recurrence are eradicated. When positioned in the right chest to reach the midthoracic esophagus, the techniques described to protect the tracheal repair remain important. Nonetheless, the need for and choice of esophageal replacement are actually a quite distinct problem that fortunately only rarely accompanies recTEF repair.

In conclusion, we have gained an increased appreciation of the need for a comprehensive and customized approach to cases of postoperative TEF. Additional complicating issues for the esophageal atresia patient are the rule, rather than the exception. Respiratory and feeding difficulties are closely interrelated and a successful treatment strategy mandates that all of these issues be evaluated and considered simultaneously. Even a brief glance at the additional procedures that have been required in this group includes: fundoplication or fundoplication revision, hiatal hernia repair, aortopexy and/or tracheopexy, laryngeal cleft repair, and esophageal replacement. Increasingly, these complex patients are likely to benefit from centralization in specialty referral centers.

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