Effect of Posterior Tracheopexy on Risk of Recurrence in Children after Recurrent Tracheo-Esophageal Fistula Repair

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BACKGROUND: A recurrent tracheo-esophageal fistula can complicate esophageal atresia and tracheo-esophageal fistula (TEF) repair in children. Therapeutic approaches and the rate of recurrence vary widely. Most reports are limited by small cohorts and short-term follow-up, and rates of re-recurrence are substantial, making it difficult to select the treatment of choice. We aimed to review our experience with the treatment of recurrent TEF using posterior tracheopexy, focusing on operative risks and long-term outcomes.

STUDY DESIGN: We conducted a retrospective review of patients with esophageal atresia TEF with recurrent TEF treated at 2 institutions from 2011 to 2020. We approach recurrent TEFs surgically. Once the TEF is divided and repaired, the membranous trachea is sutured to the anterior longitudinal ligament of the spine (posterior tracheopexy) and the esophagus is rotated into the right chest (rotational esophagoplasty), separating the suture lines widely. To detect re-recurrence, patients undergo endoscopic surveillance during follow-up.

RESULTS: Sixty-two patients with a recurrent TEF were surgically treated (posterior tracheopexy/rotational esophagoplasty) at a median age of 14 months. All had significant respiratory symptoms. On referral, 24 had earlier failed endoscopic and/or surgical attempts at repair. Twenty-nine required a concomitant esophageal anastomotic stricturoplasty or stricture resection. Postoperative morbidity included 3 esophageal leaks, and 1 transient vocal cord dysfunction. We have identified no recurrences, with a median follow-up of 2.5 years, and all symptoms have resolved.

CONCLUSIONS: The surgical treatment of recurrent TEFs that incorporates a posterior tracheopexy and rotational esophagoplasty is highly effective for preventing re-recurrence with low perioperative morbidity. (J Am Coll Surg 2021;232:690−698. © 2021 by the American College of Surgeons. Published by Elsevier Inc. All rights reserved.)

Recurrence of a tracheo-esophageal fistula (TEF) is reported in up to 10% to 15% of infants and children after repair of an esophageal atresia (EA) and TEF.[1-9] The term *recurrent TEF* is often used broadly in all patients with a postoperative fistula between the esophagus and airway regardless of the location or the etiology of the fistula.

CME questions for this article available at http://jacscme.facs.org

Disclosure Information: Authors have nothing to disclose. Timothy J Eberlein, Editor-in-Chief, has nothing to disclose. Ronald J Weigel, CME Editor, has nothing to disclose.

Drs Kamran and Zendejas contributed equally to this work. Drs Hamilton and Jennings co-senior authored this article.

Presented virtually at the 2020 American Pediatric Surgical Association (APSA) Annual Meeting and the 2020 American Academy of Pediatrics (AAP) National Conference.

Received December 16, 2020; Revised January 14, 2021; Accepted January 14, 2021.

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https://doi.org/10.1016/j.jamcollsurg.2021.01.011
ISSN 1072-7515/21

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However, precise categorization is critical when evaluating postoperative TEFs, as they each present their own diagnostic and treatment challenges. We have previously proposed that postoperative TEFs should be classified as "truly" recurrent to the site of the original fistula, missed congenital, or acquired along a new pathway—often from operative or endoscopic injury (tracheo-esophageal, esophago-pulmonary, or esophago-bronchial).[10] Although patients can present with more than 2 types of postoperative TEF, a single recurrent TEF is the most common.[10]

Recurrent TEFs rarely close spontaneously and typically require intervention, either surgical or endoscopic. Given the perceived risks of reoperative surgical intervention, many recurrent TEFs are first approached via endoscopic means with therapies that range from cauterization to injection of adhesive, sclerosant, or irritating substances into the fistula.[5-9] Unfortunately, these efforts often require multiple interventions, and the reported re-recurrence rates for endoscopic interventions remain high, ranging from 45% to 63% in small patient series with short-term follow-up.[5-9] Historically, reoperative surgical interventions have sought to separate the repaired fistula and place vascularized tissue (eg muscle or pleura) or prosthetic material (mesh) between the closed fistula to prevent re-recurrence.[11-13] Unfortunately, published results of surgical interventions are also limited to small series and short-term follow-up, and re-recurrence rates are better than with endoscopic interventions; postsurgical recurrence rates still range from 11% to 22%.[5,6,9] Although interposing tissue between the trachea and esophagus to prevent re-recurrence after a recurrent TEF repair is intuitive, it has the potential to create excessive scarring and mass effect that can compromise the airway and/or esophagus, and manifest itself clinically as extrinsic tracheal compression or refractory esophageal stricture.

Previously, we described performing a posterior tracheopexy along with a rotational esophagoplasty as part of recurrent TEF repair to separate the fistula repair suture lines without the need to interpose tissue.[10] Our results were excellent with no recurrences; however, that study addressed a variety of postoperative TEF types, including missed congenital and acquired TEFs, and only a portion of the patients underwent posterior tracheopexy, a technique that was devised in the middle of that study period. For this study, we aimed to examine our experience with the treatment of purely recurrent TEFs using posterior tracheopexy as part of the repair, focusing on their operative risks and long-term outcomes.

METHODS

Study design

With IRB approval, a retrospective review was conducted of all EA-TEF patients with a recurrent TEF who were treated at the Esophageal and Airway Treatment Center at Boston Children's Hospital between October 2011 and April 2020. Similar patients treated at Johns Hopkins All Children's Hospital in Florida from August 2019 to July 2020 by one of our former surgeons (CJS) were also included. Data collected included patient demographics, original type of EA-TEF lesion according to the Gross classification, earlier attempted endoscopic or surgical repair, operative and postoperative variables, and long-term follow-up clinical and endoscopic data.

Preoperative evaluation

Our evaluation for all patients suspected of having a postoperative TEF includes a thorough gastrointestinal and airway assessment using flexible esophagogastroduodenoscopy with fluoroscopic contrast esophagram, as well as diagnostic laryngoscopy and dynamic rigid 3-phase tracheobronchoscopy.[14,15] Our multidisciplinary evaluation is designed to determine the presence and anatomic location of the postoperative TEF, as well as identify synchronous esophageal and airway lesions, such as an esophageal anastomotic stricture or leak, acquired or missed congenital TEFs, supraglottic issues, laryngeal cleft, vocal cord dysfunction, subglottic pathology, tracheal diverticula, airway compression, and tracheobronchomalacia. Our preference is to correct concurrent esophageal and airway issues at the time of the repair of the recurrent TEF, if possible.

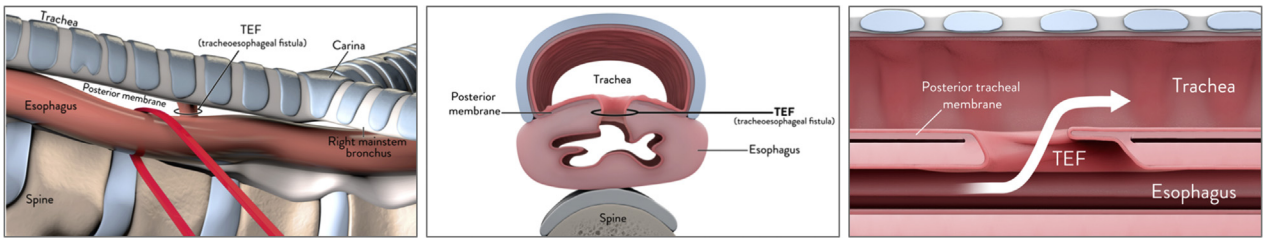

Figure 1. Recurrent tracheo-esophageal fistula (TEF) and its anatomic relationships with the trachea, esophagus, and spine. (Reprinted courtesy of the artist, Will McAbee).

Surgical technique

The surgical approach is typically through a posterior muscle-sparing thoracotomy (often right-sided) with or without a cervical incision for fistulas located in the cervical or thoracic inlet region. The interspace entry point is chosen depending on the location of the fistula on preoperative imaging; occasionally, entry via more than 1 interspace is needed. The lung is carefully mobilized from the chest wall and posterior mediastinum, taking care to protect the thoracic duct and the recurrent laryngeal and vagus nerves from injury. Our current practice is to perform recurrent laryngeal nerve monitoring for all cases with endotracheal tube surface electrodes (Dragonfly; Neurovision Medical Products) or NIM (Medtronic Inc) with or without continuous vagal nerve stimulation (APS; Medtronic Inc). The esophagus above and below the fistula is completely separated from the trachea to reveal the recurrent TEF (Fig. 1). We do not place catheters or wires endoscopically through the fistula to help find the fistula as we mobilize the entire esophagus off the airway, and the fistula site is usually readily apparent with this approach. In some cases, simultaneous esophagogastroduodenoscopy can be a useful adjunct to identify the recurrent TEF.

Once the fistula is identified, we divide it first at the esophageal level, leaving a ligated stump or diverticulum on the tracheal side (Fig. 2A). The residual tracheal diverticulum is dissected fully into the airway with the goal to remove all of the redundant mucosal lining of the fistula/diverticula and create a repair on the posterior tracheal membrane that is flush with the tracheal wall (Figs. 2B-2E). To do so, one must achieve a dissection plane that goes directly on the mucosal wall of the diverticulum—particularly at the caudal aspect of the diverticulum, as the entry point of the fistula into the airway is often slanted in a diagonal intramural path and not straight into the airway as one might expect based on the external appearance of the fistula. This dissection is guided by flexible intraoperative bronchoscopy. One or 2 safety sutures are placed (but not tied) before entering the tracheal lumen in the orientation of the planned closure of the tracheal defect (often transversely), such that once entry into the trachea occurs, one can maintain control of the airway by crossing the safety sutures. If the fistula location is proximal to the mid-trachea, one can facilitate this step by advancing the cuff of the endotracheal tube beyond the site of the fistula to reduce the air leak at the time of the fistula repair. Once all of the fistula/diverticular mucosa is dissected off and removed from the inner tracheal ridge that is located on the caudal aspect of the airway entry point, additional sutures of fine absorbable monofilament (PDS; Johnson & Johnson Medical NV) are placed to repair the tracheal defect, striving to invert or roll the tracheal mucosa inward with each stitch. Once the tracheal repair is complete, an air leak test is performed by evaluating the repair site under water with transient sustained ventilation pressure of 40 mmHg.

Subsequently, a posterior tracheopexy (regardless of the presence or absence of tracheomalacia) that includes the site of the fistula repair along with any other tracheal or bronchial areas with malacia are pexied to the anterior longitudinal ligament of the spine (Fig. 3A).[15-17] If the malacia extends to the left bronchus, a posterolateral descending aortopexy might need to be considered.[18] The esophagus is then repaired according to the magnitude of the esophageal defect and coexisting pathology. Often the esophageal defect is small and readily closed with a few sutures. However, if there is an associated stricture at the site of the fistula, a Heineke-Mikulicz strictur-oplasty or segmental esophageal resection can be performed. Once the esophagus is repaired, it is evaluated endoscopically for patency, and an air leak test is also performed under water. Esophageal perfusion is assessed with indocyanine green and SPY Portable Handheld Imager Fluorescence Imaging Technology (Stryker Corp). The esophageal suture line is rotated away from the trachea, a process that is facilitated by closing the posterior mediastinal pleura (when available) between the esophagus and the trachea, effectively keeping the esophagus intrapleural (rotational esophagoplasty, Fig. 3B).

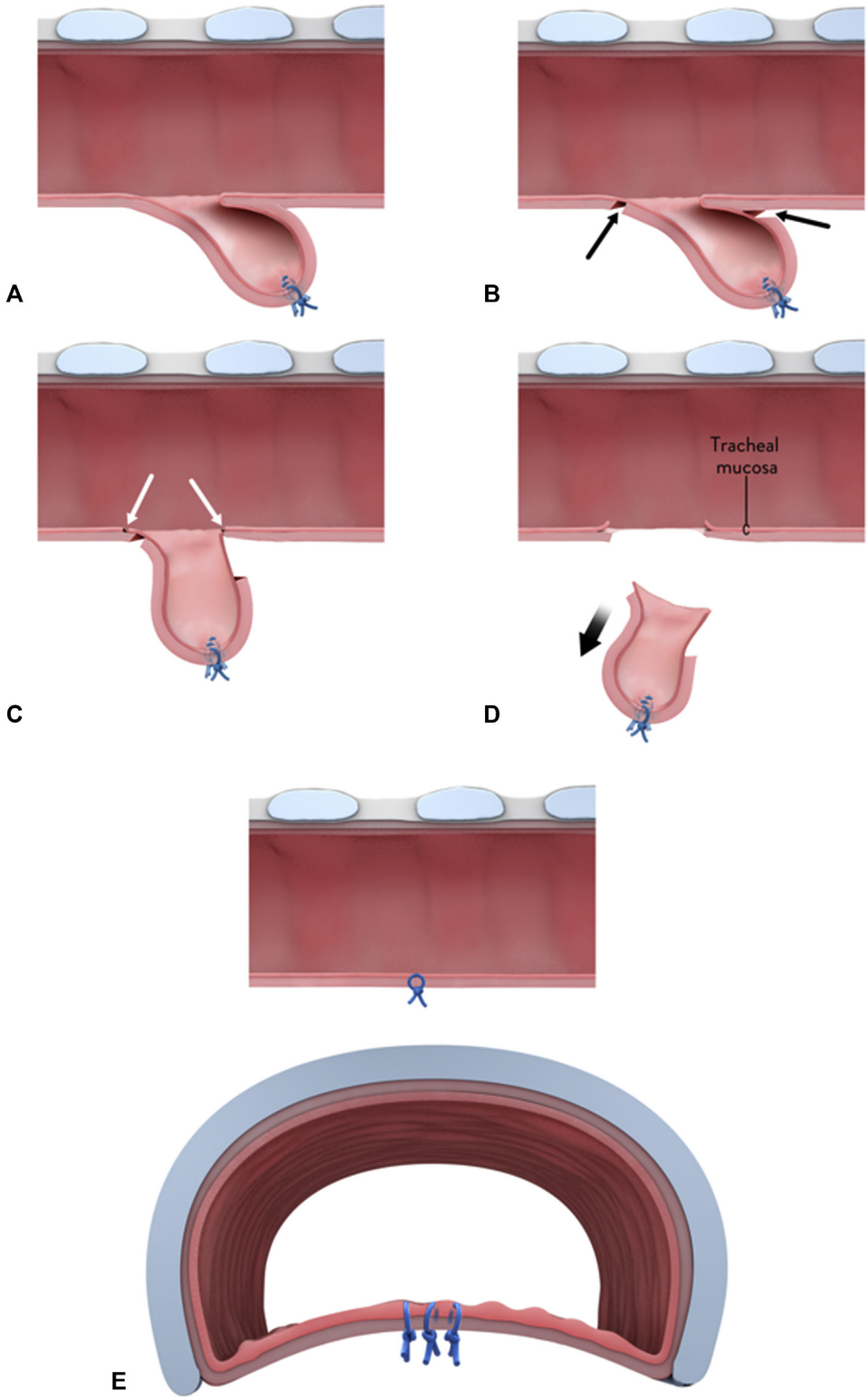

Figure 2. Resection of recurrent tracheo-esophageal fistula. (A) The fistula is divided at the esophageal level, leaving a ligated diverticulum on the tracheal side. (B, C, D) The residual tracheal diverticulum is dissected along the submucosal plane up to the entrance to the airway, removing the redundant mucosal lining of the diverticulum and leaving a flush repair on the posterior tracheal membrane. (E) Closure of the tracheal defect, rolling the mucosa inward. (Reprinted courtesy of the artist, Will McAbee).

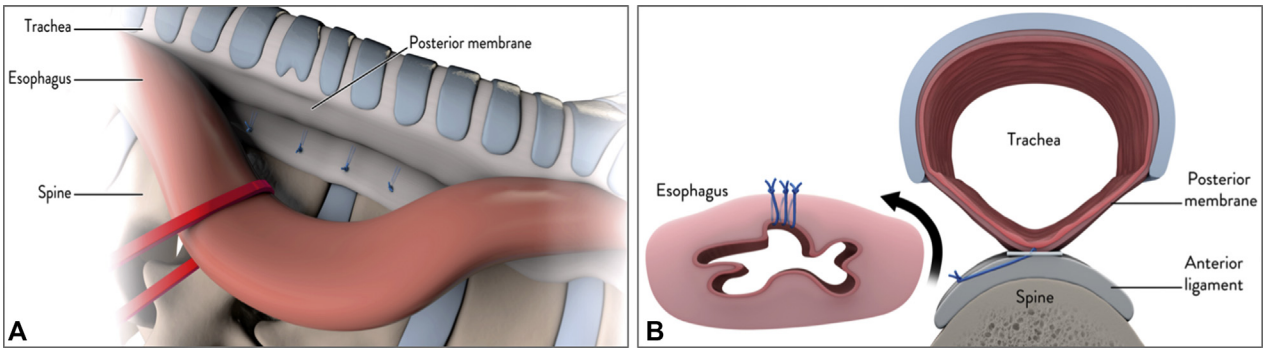

Figure 3. (A) Posterior tracheopexy: the membranous trachea at the site of fistula repair and areas with tracheomalacia sutured to the anterior spinal ligament. (B) Rotational esophagoplasty: the esophagus is mobilized to the right and the suture line rotated away from the trachea, further separating the suture lines of the fistula repair. (Reprinted courtesy of the artist, Will McAbee).

Postoperative follow-up

Patients are cared for postoperatively initially in the ICU. We strive to extubate patients as soon as possible. A postoperative contrast esophagogram is obtained 1 week after operation; oral and/or enteric feeds are then resumed if no leak is identified. All patients undergo endoscopic surveillance with fluoroscopic contrast esophagogram and rigid tracheobronchoscopy. The timing of their first endoscopic surveillance varies; if a complete esophageal anastomosis was performed, we typically assess the repair endoscopically at 1 month postoperatively. If the esophageal repair was minimal, we evaluate yearly for the first few years, and then as clinically indicated. Vocal cord function is evaluated in all patients with a flexible nasolaryngoscopy exam at the bedside or in clinic.

Table 1. Demographics and Preoperative Characteristics of Patients with a Recurrent Tracheo-Esophageal Fistula Who Underwent Surgical Repair

	Patients (n = 62)	
Variable	**n**	**%**
Sex, m	31	50
EA-TEF type (Gross classification)		
B	1	2
C	58	93
D	2	3
E (H type)	1	2
Index EA-TEF repair (elsewhere)	60	97
Original repair approach		
Open operation	59	95
Thoracoscopy	3*	5
Earlier attempted repair of recurrent TEF (elsewhere)	24	39
Open surgical	15	24
Endoscopic	13	21

*One converted to open.
EA, esophageal atresia; TEF, tracheo-esophageal fistula.

Statistical analyses

Descriptive or summary statistics are provided, with continuous variables reported as mean and SD if normally distributed or as median and interquartile range (IQR) if non-normally distributed. Categorical variables are reported as frequencies and percentages. Microsoft Excel software was used for summary statistics.

RESULTS

Between October 2011 and July 2020, sixty-two patients (59 from Boston Children's Hospital and 3 from at Johns Hopkins All Children's Hospital) with a recurrent TEF underwent surgical repair using the technique described. The majority had a history of EA-TEF Gross type C (n = 58, Table 1). All patients were symptomatic. The most common presenting symptoms were respiratory in nature, including coughing, choking, increased respiratory secretions, and recurrent pneumonia. The majority of the patients (n = 60) had their index TEF repair at a referring institution, predominantly via thoracotomy (n = 59). Most (n = 59) had undergone a primary esophageal anastomosis for their EA repair, 1 required a reversed gastric tube and 2 patients presented as unrepaired EA after an initial TEF repair that recurred. Twenty-four patients (39%) had at least 1 previous surgical (24%) and/or endoscopic (21%) attempt at recurrent TEF repair. Endoscopic methods included TEF mucosal injury by laser, sclerosis, or cautery; use of tissue sealants, such as fibrin glue and Histoacry (TissueSeal); or placement of an endovascular plug device. Most patients with an earlier surgical attempt at recurrent TEF repair had muscle, pleura, or biologic mesh interposed between the TEF repair sites.

At the time of the recurrent TEF repair at our institutions, patients' median age was 14 months (IQR 7 to 21 months) and median weight was 8 kg (IQR 6.4 to 12.5

Table 2. Operative Details of Patients with a Recurrent Tracheo-Esophageal Fistula Who Underwent Surgical Repair

Variable	Patients (n = 62)
Age at time of surgical repair, mo, median (IQR)	14 (7–21)
Weight at time of surgical repair, kg, median (IQR)	8 (6.4–12.5)
Surgical approach, n (%)	
Thoracotomy	
Right	58 (93)
Left	1 (2)
Cervical incision	2 (3)
Robot-assisted thoracoscopy	1 (2)
Mediastinal cyst/abscess resection, n (%)	9 (15)
Esophageal stricture segmental resection, n (%)	11 (18)
Esophageal stricturoplasty, n (%)	18 (29)

IQR, interquartile range.

kg) (Table 2). Most patients (n = 59) underwent repair of their recurrent TEF by re-do thoracotomy (right-sided except in 1 case), 2 by cervical incision, and 1 by robot-assisted thoracoscopy. Nine patients (15%) also required resection of a large mediastinal cyst or abscess that was related to the tracheal diverticulum (likely resulting from an earlier double ligation of the TEF) (Fig. 4). For esophageal repair, most patients (n = 33) had no associated esophageal stricture, and the quality of the esophageal tissue was adequate to undergo simple suture repair after debridement of the TEF site back to healthy edges. For those with a concomitant esophageal anastomotic stricture (n = 29), which was almost always in close proximity to the TEF site, the esophageal repair was guided by the length and severity of the stricture as assessed by endoscopic and operative means; some required a Heineke-Mikulicz type stricturoplasty (n = 18), and others required a segmental esophageal stricture resection (n = 11). Of note, in 2 patients, the stricture length and tissue quality were such that after stricture resection, it was not possible to anastomose the esophagus primarily. Therefore, these 2 patients underwent external traction-induced esophageal lengthening process (Foker procedure).

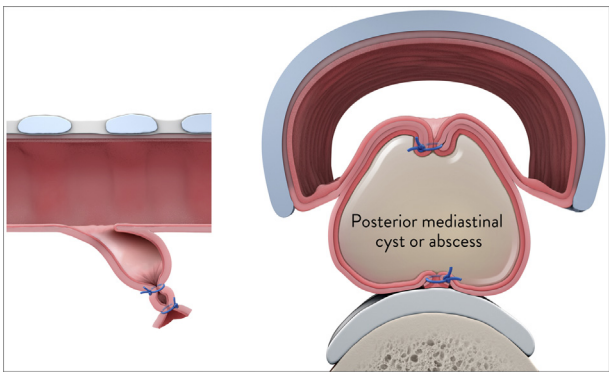

Figure 4. Mediastinal cyst/abscess related to the tracheal diverticulum, likely resulting from an earlier double ligation of the tracheo-esophageal fistula. Note its potential for airway intrusion. (Reprinted courtesy of the artist, Will McAbee).

Nearly all of our patients have returned for clinical and endoscopic follow-up, leading to a median follow-up of 2.5 years (IQR 1 to 5 years). Unfortunately, 5 patients from overseas were lost to follow-up. Nonetheless, all patients have undergone at least 1 postoperative endoscopic airway and/or esophageal evaluation (including fluoroscopic contrast esophagram); their initial endoscopy was performed at a median of 3 weeks (IQR 2 to 4 weeks) after recurrent TEF repair. Not a single patient has been identified to have a re-recurrent TEF (Table 3). All patients report resolution of their respiratory symptoms.

Perioperative morbidity

Esophageal anastomotic leak developed in 3 patients (5%); 2 patients healed with nonoperative management and a chronic leak developed in the third patient after a Foker process and esophageal replacement was ultimately required. Two additional patients required an esophageal replacement. One of them secondary to a refractory stricture after a Foker process, and the other from multiple acquired esophagopulmonary fistulas. Mild esophageal strictures developed in 17 patients (27%) who responded favorably to endoscopic anastomotic dilations at their follow-up endoscopies. Ten patients (17%) had preexisting unilateral vocal cord dysfunction, of which only 2 have resolved. One patient (2%) experienced new unilateral vocal cord dysfunction after repair of the recurrent TEF, but this resolved on repeat examination. There were 2 deaths (4%). One was from operative hemorrhage related to an aortic injury at the time of recurrent TEF repair. This patient, with a right-sided aortic arch and multiple previous failed endoscopic attempts at recurrent TEF repair, had significant peri-aortic inflammation causing the fistula to adhere to the aorta. The patient survived the immediate operation after aortic patch repair on cardiopulmonary bypass, but had an irreversible neurologic injury. The second patient with multiple medical comorbidities died from his underlying cardiac disease several months after the recurrent TEF repair.

DISCUSSION

This study represents the largest reported experience to date with the operative treatment of patients with recurrent TEFs. In a field where re-recurrent TEFs are common, the findings of this study demonstrate that it is possible to achieve a negligible long-term recurrence rate. We

Table 3. Postoperative Outcomes of Patients with a Recurrent Tracheo-Esophageal Fistula Who Underwent Surgical Repair

Variable	Patients (n = 62)
Follow-up interval, y, median (IQR)	2.5 (1−5)
Re-recurrence of TEF, n	0
Resolution of respiratory symptoms, n (%)	62 (100)
Postoperative esophageal anastomotic dilation, n (%)	17 (27)
No. of dilations per patient, median (IQR)	2 (1−5)
Esophageal anastomotic leak,* n (%)	3 (5)
Esophageal replacement by jejunal interposition,† n (%)	3 (5)
Vocal cord paralysis (new), n (%)	1 (2)
Death,‡ n (%)	2 (3)

*Two anastomotic leaks were treated nonoperatively and healed, and a chronic leak developed in 1 patient after a failed Foker process and esophageal replacement was eventually needed.
†Two patients required esophageal replacement after failed Foker processes (one from a chronic leak as above and the other from dysmotility and a chronic stricture), and the third patient required esophageal replacement due to development of multiple acquired esophago-pulmonary fistulas.
‡One unrelated to the recurrent TEF repair.
IQR, interquartile range; TEF, tracheo-esophageal fistula.

therefore advocate that complete mobilization of the esophagus from the trachea with division of the recurrent TEF and separation of the suture lines by posterior tracheopexy should be considered the operative procedure of choice for recurrent TEF. The primary reason to attempt a less invasive endoscopic approach to treat a recurrent TEF has been the perceived morbidity that occurs with an operative repair. Although some patients with recurrent TEF might be treated successfully with an endoscopic intervention, it is clear from the data published in the literature that the risk of re-recurrence is fairly high.[5-9] In addition, even if successful, endoscopic treatments address the recurrent TEF only, which is often not the only problem these patients face. With a surgical approach, the benefits of addressing co-existent tracheomalacia, esophageal stricture, and other associated intra-thoracic pathology are tangible. We have also seen that repeated endoscopic attempts can be futile and/or harmful, and can make the eventual operation harder by creating increased local tissue scarring and/or damage. Therefore, in our practice, an upfront surgical approach is preferred. However, we recognize that in centers without the expertise or resources to carry out this operation safely, an endoscopic approach might be considered. Given that not all recurrent TEFs are similar in length, width, or location, additional research is needed to examine which recurrent TEF characteristics are associated with greater likelihood of endoscopic success. Still, surgeons should be cognizant of the diminishing prospect of success after repeated endoscopic failures, the potential risk of multiple anesthetics to the developing brain of the child,[19-23] and ongoing pulmonary morbidity from chronic aspiration from an untreated recurrent TEF, to timely refer the child for a definitive operation.

Although the frequency of operative morbidity and mortality in our series is low overall, some of these complications can be considerable. One must be particularly cautious in cases that could be at increased risk of complications developing, such as those requiring a long-segment esophageal stricture resection. In these situations, one should have a backup esophageal reconstruction plan in place (Foker process or esophageal replacement) in the event that the stricture resection results in a gap that is not amenable to primary repair. In this setting, the choice of esophageal reconstruction is often highly dependent on locally available expertise and resources. Although the 2 patients who underwent a Foker procedure in our series ultimately required an esophageal replacement, in our practice, we believe that whenever there is residual healthy esophagus, attempting to preserve the native esophagus is generally a better option than an upfront esophageal replacement. Even though the success rate with the Foker process strategy is not perfect, we have been able to save several children from needing an esophageal replacement.[24] In addition, patients with poor cardiopulmonary reserve, aberrant great vessel anatomy, and/or substantial peri-aortic inflammation are also at increased perioperative risk, for which early involvement of a cardiac surgical team for pre-emptive cardiac bypass or extracorporeal membrane oxygenation cannulation should be considered to create a safer environment for a challenging posterior mediastinal dissection.

The identification of recurrent TEFs can be challenging, as many of these are very subtle.[25-27] Although rigid bronchoscopy has been considered the reference standard for diagnosis of a TEF, we have previously shown that a combination of techniques that incorporate flexible esophagoscopy with contrast esophagram, end-tidal CO_2 monitoring, and a thorough airway assessment with rigid and/or flexible tracheobronchoscopy augment the sensitivity and diagnostic accuracy of the evaluation.[14] We strongly believe that a careful multisystem and multidisciplinary preoperative evaluation is essential to identify the number, location, and character of the postoperative TEFs along with coexistent airway, esophageal and/or intra-thoracic pathology to achieve low operative morbidity with these procedures. Similarly, the patient's nutritional status and cardiopulmonary physiology should be optimized preoperatively.

Given the effectiveness of a posterior tracheopexy and rotational esophagoplasty to prevent re-recurrence of a TEF, one could envision its potential role in the primary prevention of a recurrent TEF. In fact, our group and others have previously described performing a posterior tracheopexy at the time of initial esophageal atresia repair in select patients.[17,28]

Clearly, additional study is needed to determine whether a posterior tracheopexy at the time of the original EA repair will decrease the incidence of recurrent TEF and the development of tracheomalacia symptoms later in life. It is also unknown what additional morbidity will occur if every infant with a TEF undergoes this additional procedure.

Acquired TEFs are often esophago-pulmonary (parenchymal) in nature; for these we recommend full separation of the fistula, followed by esophageal and parenchymal lung repair. The challenge then becomes trying to separate the lung parenchymal repair site from the esophageal repair site. Options include a second layer of parenchymal lung repair that imbricates the original repair, parietal pleural patch (if available), and a rotational esophagoplasty placing the site of the esophageal repair against the spine or chest wall.

Our study has several important limitations. Although our robust follow-up and the thoroughness of our postoperative endoscopic evaluations are well beyond what has been reported previously, 5 international patients who were lost to follow-up could have had a recurrence develop, of which we are not aware. In addition, given that our operative technique incorporates several elements (ie posterior tracheopexy and rotational esophagoplasty), it is not possible to fully determine which of the elements is responsible for the efficacy of the repair, although we believe it is the combination of all elements. In addition, data collection was retrospective and follow-up was gleaned from the chart review. Future efforts will entail active contact of our cohort to evaluate the quality of life and other patient-reported outcomes metrics that these patients experience as a result of our intervention, such as chest wall deformities from repeated thoracotomies.[29] Lastly, although we are able to show that one of our former colleagues was able to replicate our technique and results at another institution, we recognize that both centers are highly specialized referral practices for children with complex esophageal and airway problems, and that our results might not be generalizable to other centers. In fact, how much of our results are a reflection of a volume to outcomes relationship remains to be studied.

CONCLUSIONS

Our results suggest that the operative treatment for a recurrent tracheo-esophageal fistula should include a full separation and repair of the esophagus and trachea along with a posterior tracheopexy and rotational esophagoplasty, as we have described in detail. With this approach, one can achieve a negligible re-recurrence rate with low perioperative morbidity and mortality. Given the rarity and complexity of infants with recurrent TEFs, they might be best treated in a center of experience with these complex issues.

Author Contributions

Study conception and design: Kamran, Zendejas, Meisner, Choi, Munoz-San Julian, Ngo, Manfredi, Yasuda, Smithers, Hamilton, Jennings

Acquisition of data: Kamran, Zendejas, Meisner, Choi, Munoz-San Julian, Ngo, Manfredi, Yasuda, Smithers, Hamilton, Jennings

Analysis and interpretation of data: Kamran, Zendejas, Smithers

Drafting of manuscript: Kamran, Zendejas

Critical revision: Kamran, Zendejas

Acknowledgment: The authors would like to express their appreciation to Dr Robert C Shamberger for his guidance and helpful advice throughout this project. The authors also thank Dori Gallagher and the multidisciplinary team of the Esophageal and Airway Treatment Center at Boston Children's Hospital.

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