

## Review of (acquired) incidental, rare and difficult tracheoesophageal fistula management

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### Abstract

Acquired benign tracheoesophageal fistula is a rare condition and a difficult problem. The rarity and unpredictable presentation of this condition makes the design and setting of randomized prospective trials impossible. Guidelines on this matter are also difficult to establish. Based on a comprehensive evaluation of published literature and their experience, the authors review the etiology and best options for treatment, either surgical and non surgical, according to present knowledge.

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**Key words:** Tracheoesophageal fistula; Esophageal stents; Tracheal stents; Surgical treatment

**Core tip:** Acquired nonmalignant tracheoesophageal fistula is a rare life-threatening condition. Several management approaches have been proposed, without a real consensual approach. The authors review the published literature and discuss the different options.

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### INTRODUCTION

Acquired benign tracheoesophageal fistula (TEF) is a rare condition and a difficult problem that simultaneously compromises the respiratory and digestive functions. Morbidity is very high and, in untreated patients, mortality is probably close to one hundred percent. Similarly, treatment is also very difficult and published collective experience scarce. The rarity and unpredictable presentation of this condition makes the design and setting of randomized prospective trials impossible and is a limiting factor for the quality of information derived from the very few retrospective series published so far. Guidelines on this matter are also difficult to establish since the few published data differ significantly in issues like fistula etiology and location and the clinical expertise of surgeons (thoracic, general, ear, nose and throat) and gastroenterologists.

Therefore, for surgeons facing this difficult issue, a full and comprehensive evaluation of the literature should consider all the published data and the specificities of the information provided, such as the correct assessment of hospital resources, namely, the collective experience of a mandatory multidisciplinary approach. In such a difficult and rare condition, to reach a large and sound clinical experience is very challenging. At best, the concurrent experience in other clinical fields will hopefully provide the skills to deal with acquired benign tracheoesophageal fistulas. Due to the complexity of this condition, a clinical surgeon uncomfortable with the management of this disease should refer these patients to an experienced center.

### THE SURGICAL APPROACH

Five important papers published on this subject can be

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identified<sup>[1-5]</sup>, coming from experienced surgical groups with a sound reputation and experience and reporting clinical good outcomes. However, none of those groups were able to treat more than 75 patients and only over a long period of 30 to 35 years could those numbers be reached. Published scientific evidence is, at best, on the expert opinion range (level 3). Hilgenberg *et al*<sup>[1]</sup> were probably the first to publish a systematic review on this complication based in their personal experience with 20 patients. Lesions were caused by tracheal intubation (14), blunt trauma (3), orthopedic cervical spine procedures (2) and foreign body ingestion (1). Almost all of these lesions involved the proximal esophagus and the surgical approach relied on tracheal resection and anastomosis with either a direct suture of the esophageal perforation (16 patients) or an end to end reconstruction (3). Mortality reached 10% and fistula recurrence 5%. The most useful recommendations were the importance of pre-operative mechanical ventilation weaning and the use of interposition of healthy muscular tissue buttressing the tracheal and esophageal suture lines.

Mathisen *et al*<sup>[2]</sup> reported their results in 1991 with a series of 38 patients treated for tracheoesophageal fistulas over a 16 year period, later completed with another 36 patients operated on from 1992 to 2010<sup>[3]</sup>. Interestingly in this series, the largest published until now, the etiology changed, with a decreasing incidence of post intubation injuries (71.1% to 47.2%) whilst other causes, like esophageal surgery and laryngectomy complications, increased in prevalence (5.3% to 27.8%). Reported fistulas were mostly located in the mid and upper trachea (61% and 36%). The majority (92%) were less than 3 cm long. Surgical approach was mostly cervical or cervical plus upper sternotomy. There was a clear trend change, from tracheal resection and anastomosis to direct and simple repair of the tracheal lesions, during the time span of this study, which the authors attributed to the increase of complications of esophageal and laryngeal surgery as the cause of tracheoesophageal fistulas. In this setting, compared with post intubation injury, the destruction of tracheal tissue was found less disruptive and more suitable for a conservative approach. Although mortality decreased from over 10% to 2.8% in the second period, fistula recurrence more than doubled, general complications remained the same, the number of patients requiring a tracheal procedure increased more than four fold, and the patients that were not able to recover oral intake were in excess of 17.1%, a five fold increase over the first time period. The authors established a relationship between these events and the minor tracheal lesions, TEF occurring after resection of the esophagus or larynx, and they considered that the later conditions were more challenging problems with a higher rate of fistula recurrence. They also reinforce the statement for the use of healthy muscular tissue to protect suture lines, underlining the importance of mechanical ventilation weaning before endeavouring tracheal reconstruction. For ventilator dependent patients, the authors emphasize the need for an adequate endotracheal tube cuff placement distal to the

fistula opening. They also sustain the need for optimization of the overall medical condition prior to any definitive surgical approach, through placement of a feeding jejunostomy and a decompression gastrostomy, the removal of nasogastric feeding tubes (which adds further damage to tissues), and control of sepsis. They argue against the use of temporary or definitive esophageal stents because, in their opinion, they do not contribute to the treatment of established lesions and may also enlarge TEF, creating giant fistulas.

Another very interesting study comes from Italy with Baisi *et al*<sup>[4]</sup> reporting 31 patients operated on for tracheoesophageal fistulas over a period of 18 years. In this series, two thirds of the fistulas were caused by endotracheal intubation. The other significant cause was orthopedic cervical spine surgery (4 patients). Laryngeal surgery was not identified as a cause and esophageal surgery accounted with only one case of a Zenker's diverticulectomy as the primary procedure. Fistulas were all proximal in the trachea and surgical approach was mainly cervical. Again, they agree with previous authors on the need for weaning the patient from mechanical ventilation and obtaining an optimal general and medical condition with endoscopic percutaneous gastrostomy, feeding jejunostomy and sepsis control. In their experience, tracheal resection and reanastomosis was rarely needed since 26 patients were treated with tracheal and esophageal direct suture. This approach is contradictory to Mathisen's claim that post intubation lesions are more disruptive of tracheal tissue and more often require tracheal resection. These last authors also emphasize the need for muscular tissue interposition. Mortality was low, with only one reported death.

A very important series comes from the Mayo Clinic in Rochester, with Deschamps<sup>[5]</sup> presenting the results from a 30-year retrospective review including 35 patients. In this series, fistula etiology differs significantly from previous data, with most TEFs related to post-esophagectomy complications, while the post-intubation lesions accounted for less than 6% of the cases. Other important differences were the presence of trauma (17.1%), mediastinal tuberculosis (14.3%), radiation therapy (5.7%) and the *de novo* reported presence of an indwelling airway or esophageal stents as a cause for TEF (11.4%). All these etiologies were previously unreported. Not surprisingly, fistula location was more widely distributed, the majority being located distally in the carina (9) and main bronchus (14). This modified the surgical approach and strategy, with most patients being operated on through a thoracotomy or a thoracotomy plus a cervicotomy or laparotomy. In some patients, segmental bronchial resection was needed. The number of TEF requiring a multistaged repair was also important (7) and reoperations for complications (esophageal leak, bleeding, recurrence of TEF and tracheal dehiscence) reached almost 22.8%. Despite those figures, mortality was only 5.7% and 29 patients (82.9%) were able to return to an oral diet. Still, a great number of patients were treated with single stage division of the fistula and direct repair of both the tracheal and esophageal defect. These authors concur with previous

reports on the importance of buttressing the suture lines and weaning the patients from mechanical ventilation, although they do not equally emphasize these procedures, particularly in cases where tracheal resection and anastomosis is not needed.

Bartels *et al*<sup>[6]</sup> presented a report on tracheobronchial lesions (including 4 TEF) exclusively as morbidity of post esophageal resections. They were more frequent with the transthoracic approach than with the transmediastinal route and all cases were evident up to one month after the original operation. Prevalence was 3.9%. Factors closely related to the occurrence of those lesions were neoadjuvant radiotherapy, extensive thoracic lymphadenectomy and dissection, as well as insufficiently drained local sepsis (mostly from anastomotic leaks). Despite this surgical group experience and expertise in Siewert's report, mortality averaged 33% and was correlated with the above risk factors. The authors found no positive contribution for fibrin glue or stents use and underscored the importance of weaning the patient from mechanical ventilation and of the use of buttressing of suture lines.

## THE CONSERVATIVE APPROACH

For many years, esophageal stenting has been used in the management of malignant and benign dysphagia and tracheoesophageal fistulas<sup>[7]</sup>. Tracheal<sup>[8]</sup> and combined (tracheal and esophageal) stenting<sup>[9-12]</sup> were also reported, including combined surgical and endoscopic approaches. The results from these studies are difficult to analyze due to the mixed nature of the pathologies involved (benign, malign, strictures, isolated esophageal or tracheal fistulas) and the diversity of stents used (plastic, metallic, covered or uncovered, retrievable or not). Major criticisms on this type of solutions for benign TEF are the low rate of fistula sealing without a real cure<sup>[13]</sup>, the unnecessary and deleterious delay of definitive treatment and the potential for further damage of already traumatized tissue<sup>[14,15]</sup>. In fact, it is unlikely that the artificial surface of an esophageal prosthesis might allow, without the natural matrix provided by natural healthy tissue (muscle or other tissue buttressing), the healing of the *pars membranosa* of the trachea, the anterior wall of the native or interponate esophagus or both. This is mostly true in a patient dependent on mechanical ventilation because positive pressure will fuel the conditions for a perpetual tracheal leak. The same holds true for tracheal prosthesis alone. In this case, despite effective sealing of the airway, the esophageal leak will be responsible for local sepsis and persistent fistula. However, we found that a tracheal prosthesis that seals the airway defect might be temporarily useful, protecting the tracheal suture and tissue buttressing during unavoidable mechanical ventilation in the post operative period<sup>[8]</sup>. Its temporary and cautious use might also correct (modulate) late tracheal stenosis after surgical procedures. Recently, we used this approach with good results on a patient successfully operated on for TEF (post tracheal intubation) that subsequently developed isolated tracheal stenosis (unpublished data).

In our personal series, we also registered 2 TEF after esophageal resection for cancer (3.1% of the esophagectomies performed) with both patients submitted to neoadjuvant radiotherapy. Both patients were operated on through a thoracic approach and both suffered from long lasting cervical anastomotic leaks. The risk factors were identical to the ones reported in the Siewert<sup>[6]</sup> series but, in these cases, the TEF presented late, at 3 and 9 months after esophagectomy and cervical anastomotic leaks closure (unpublished data). A conservative approach was initially selected, with esophageal or tracheal prosthesis, but this approach failed and both patients were later operated on (tracheal and esophageal suture and sternocleidomastoid muscle interposition). One recovered uneventfully from the surgical procedure. The other patient suffered from recurrence of the fistula, reoperation, and finally, transsternal definitive tracheostomy followed by death from sepsis and multiple organ failure.

Finally, a 5<sup>th</sup> patient was operated on with a TEF resulting from a long lasting (1 year) tracheal stent initially inserted to treat a post intubation stenosis. This case underlines the indwelling esophageal or tracheal prosthesis risk of TEF.

## CONCLUSION

Treating benign TEF is challenging and a very difficult problem due to the potential devastating complications, patient suffering and death. Personal or institutional experience is scarce and even "high volume" centers face this problem at most once a year. There are no randomized studies or guidelines and only expert opinion is available<sup>[1-6,16]</sup>. Furthermore, published series differ significantly over important issues like fistula etiology and location, hospital resources and specificities of surgical and gastroenterology training. Therefore, for the occasional surgeon facing this problem, there are "off the shelf" solutions. Thus, these cases should be referred to experienced centers.

TEF patients require a multidisciplinary approach, encompassing the cooperation not only of surgical specialties (general, thoracic, ear, nose and throat), but also anesthesiologists and intensivists who in the end will have to manage and secure the airway in a complicated and difficult acute setting. This is a very important statement and only Baisi *et al*<sup>[4]</sup> report briefly and incompletely state this need. There are in fact a few studies published by anesthesiologists<sup>[17,18]</sup> reporting the difficulties they faced and the imaginative solutions that they used to overcome these uncommon situations. Some of these reports deserve to be carefully consulted, discussed and made available to all surgical teams as in some cases the reported "tricks" may make a substantial difference.

From the surgical point of view, some important issues are consensual. Almost all groups agree on the advantage of unsupported ventilation before any major surgical procedure. An optimal medical condition also should be pursued, namely through a gastric decompression and feeding jejunostomy tube placement. If at all

possible, the simpler surgical solution is certainly the best, that is, use of a single surgical approach (cervical or thoracic), a direct suture of the tracheal and esophageal lesions and the placement muscle interposition between suture lines. In fact, only Camargo *et al.*<sup>[19]</sup> seems to minimize the importance of this simple, harmless and effective step. In spite of the complexity and etiology of TEF, a recent trend for less frequent tracheal resections, less frequent use of multistage procedures and esophageal exclusion or diversion is apparent.

Every surgeon must be prepared for complex and demanding procedures like tracheal resection and reconstruction, laryngotracheal resection and reconstruction eventually associated with major esophageal surgery.

The use of stents in benign situations must be cautious, temporary, tailored for specific situations, and should not be considered as a definitive approach. However, during the post operative period when a distal to the suture line tracheal tube placement is not possible, they may have a role as an adjunct, either as a short bridge for a definite surgical approach or as an airway protection procedure in a mechanical ventilation dependent patient.

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