

## Repair of Congenital Tracheoesophageal Fistula Recurrence in an Adult Intensive Care Unit Patient Hospitalized due to Hemorrhagic Stroke – A Case Report and Literature Review

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### Abstract

Recurrence of tracheoesophageal fistula is a complication, characterized by high mortality risk. It may occur either directly postoperatively or several years after the initial repair. Direct closure of both tracheal and esophageal defects with the interposition of a muscle flap is the most usual surgical treatment performed. We present the case of a 65-year-old patient who was operated as an infant for a congenital tracheoesophageal fistula through a right-sided thoracotomy presenting now with a recurrent tracheoesophageal fistula caused by prolonged intubation due to a cerebral hemorrhage.

**Keywords:** Tracheoesophageal fistula; Recurrence; Surgical therapy

### Glossary of Abbreviations:

TEF: Tracheoesophageal Fistulas; rTEF: Recurrence of Tracheoesophageal Fistulas, ICU: Intensive Care Unit; EGD: Esophago-Gastro-Duodenoscopy; FTSG: Full Thickness Skin Graft; CT: Computed Tomography

### Introduction

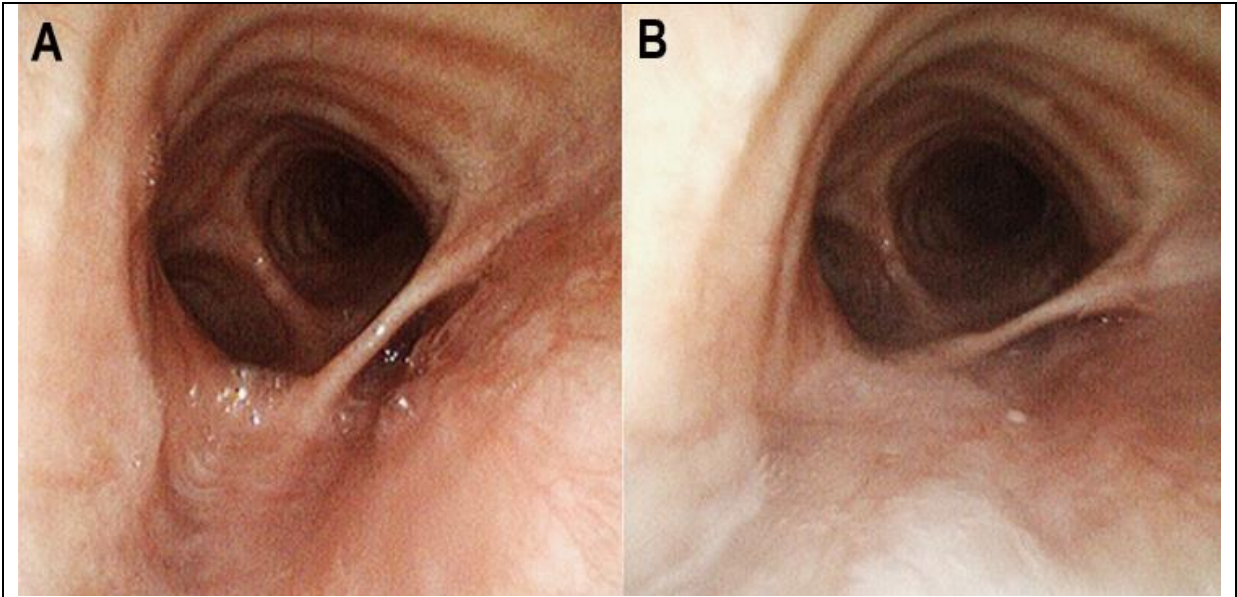
Recurrence of Tracheoesophageal Fistulas (rTEF) is a rather unusual complication characterized by high mortality, usually presenting with symptoms such as choking, cough, cyanosis with feeding and/or recurrent respiratory infections [1]. The recurrence rate of Tracheoesophageal Fistulas (TEF) after initial repair has been noted to be between 5 and 10% of cases, commonly occurred within 2-18 months after the initial repair [2]. The

success of surgery depends on several factors such as the medical history of diabetes, the presence of infection, the nutritional status of the patient, the usage of steroids and the surgeon's skills [3]. Division and closure of the fistula followed by placement of viable tissue between the tracheal and esophageal defects via thoracotomy has been historically the standard of care for rTEF [4]. Due to high morbidity and mortality rates of these techniques, less invasive interventions such as various endoscopic techniques have been introduced over the years with various success rates [2].

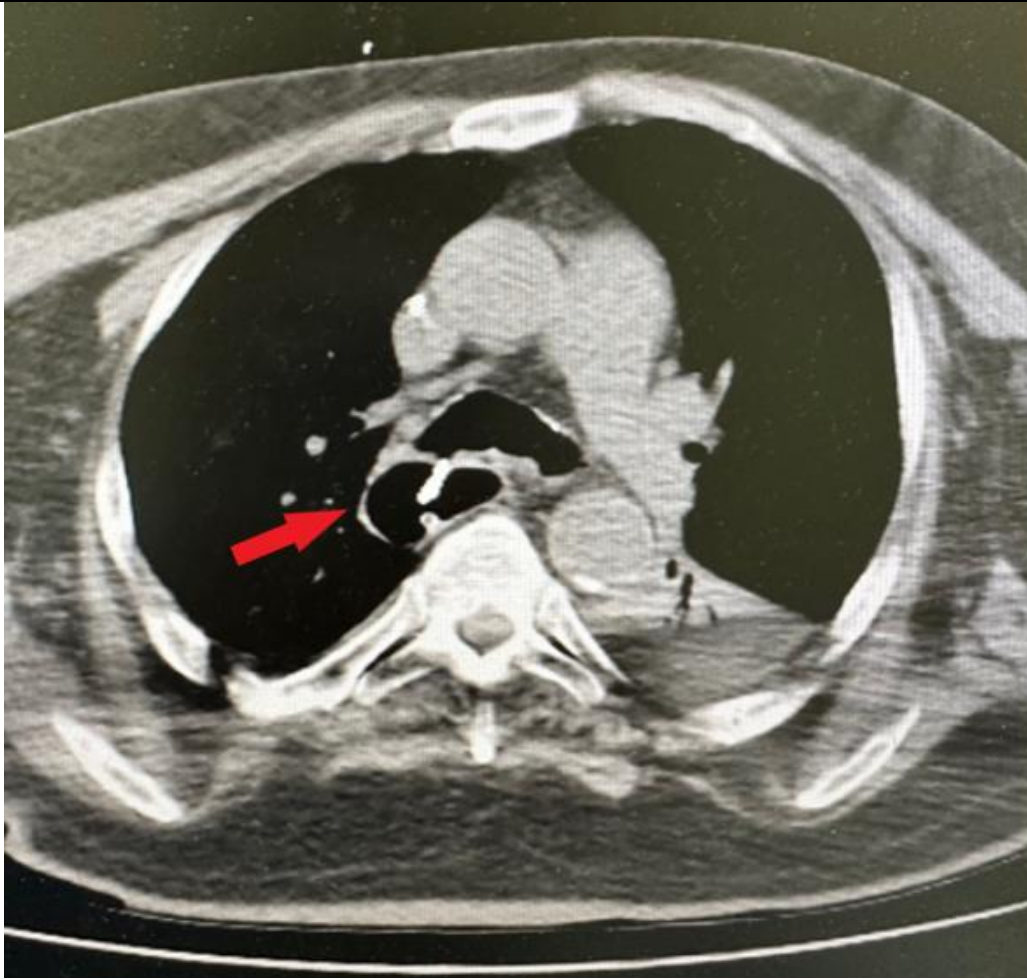
### **Case Presentation**

We present the case of a 65-year-old Caucasian man with a medical history of arterial hypertension and insulin-dependent diabetes mellitus type II, who was operated as an infant for a congenital tracheoesophageal fistula through a right-sided thoracotomy. The patient was admitted to the hospital due to an extensive cerebral hemorrhage, which required intubation and mechanical ventilation in the Intensive Care Unit (ICU). A recurrent tracheoesophageal fistula was suspected in the ICU after the observation of significant air leak during mechanical ventilation. The amount of the air leak was noticed to be dependent on the tube

position. Urgent bedside bronchoscopy (**Figure 1A**) revealed a defect of the posterior tracheal wall, 3 centimeters above the carina. The subsequent Esophago-Gastro-Duodenoscopy (EGD) confirmed the tracheoesophageal fistula, and an effort for fistula closure with clips has been done by the gastroenterologists. Air leak during mechanical ventilation continued and three days later, a Computed Tomography (CT) of the chest was performed, confirming the already known TEF between the posterior wall of the trachea and the anterior wall of the esophagus (**Figure 2**). Surgical repair of the fistula was therefore decided. During the operation extended symphysiolysis was performed due to dense adhesions caused by the periesophageal and peritracheal inflammation. The fistula was found and divided. The tracheal defect was closed with interrupted non-absorbable sutures, and the esophageal defect was closed with interrupted absorbable sutures. Lastly, a muscle pedicle graft (serratus anterior) was interposed and sutured between the trachea and the esophagus. Postoperatively, no air leak during mechanical ventilation was noticed, and the subsequent bronchoscopy confirmed the successful repair of the fistula (**Figure 1B**).



**Figure 1:** **A-**The initial bronchoscopy revealing the tracheoesophageal fistula between the posterior walls of the trachea, 3 centimeters above the carina, and the anterior wall of the esophagus. **B-**Postoperative bronchoscopy revealing successful repair of the fistula.



**Figure 2:** The initial CT scan showing the tracheoesophageal fistula and the clips placed during the EGD (red arrow).

## Discussion

A systematic review by Aworanti et al in 2013 showed that open repair had a higher success rate and required fewer treatments compared with the endoscopic therapy. In 108 patients who underwent open repair, 93,5% had successful closure of the fistula with an average of 1,1 procedures (range 1-2) and refistulization rate of 21%, whereas 57 patients who underwent endoscopic procedures had 84% success rate and 63% refistulization rate [5]. A recent review in 2023 by Tobia A, et al regarding endoscopic treatment in pediatric patients with rTEF including English, Spanish and German articles was performed. In their review the de-epithelialization techniques had the highest overall

success rate but repeated endoscopic treatment attempts were required to obtain complete closure of the fistula (there was a relatively low success rate after a single procedure) (Table 1) [2]. A few patients were treated with tracheal stents with unfavorable results [2]. Regarding surgical treatment, a cervical approach must be used in patients with a proximal fistula while a right-sided thoracotomy can be used in patients with a distal fistula [1]. Concerning surgical techniques, the method most often performed is the direct closure of both the tracheal and esophageal defects with or without pedicled muscle flaps, segmental tracheal resection, and anastomosis together with esophageal closure and/or esophageal diversion [6]. In rTEF, surgical repair is very important to protect

the esophageal suture line with a strap muscle to avoid a new recurrence [6]. Altorjay et al. recommend the dissection of a free tracheoesophageal space at the level of the bifurcation, when a distal third tracheal lesion extending to the bifurcation is present, through a right-sided posterolateral thoracotomy because at this point the membranous part of the trachea can be separated from the esophagus more safely [7]. Battistella et al. present the usage of a synthetic bioabsorbable patch to cover the defect and create a scaffold for epithelial colonization of the neomucosa and thus to guarantee a rigid support for

the intercostal muscle, reducing the risk of bulging into the tracheal lumen [3]. Huang et al. report the interposition of a patch of Full Thickness Skin Graft (FTSG) between the esophageal suture line and the tracheal wall, which acts as a mechanical barrier, hindering the passage of saliva into the trachea through the esophageal leak [6]. Lastly, Bardini et al. describe the interposition of a patch of Gore-Tex membrane between the esophageal suture line and the tracheal wall, which also acts as a mechanical barrier, allowing the spontaneous healing of both the esophageal and tracheal leaks, preventing further pulmonary complications [8].

**Table 1:** Summary of outcomes, including all case reports and case series of endoscopic treatment for rTEF.

	<b>Sealant injection</b>	<b>De-epithelialization technique</b>	<b>De-epithelialization technique + Sealant injection</b>
Number of patients treated	21	46	47
Successful single treatments, number (%)	5 (23.8%)	11(31.4%)	13 (27.7%)
Overall success rate, number (%)	9 (42.8%)	39 (84,7%)	32 (68%)

## Conclusion

The recurrence of TEF usually occurs early after the initial repair and is associated with high mortality rates. In our case the recurrence happened more than 60 years after the initial repair, which is unusual and makes this case extremely rare. According to the published literature, endoscopic treatment is a minimally invasive technique with less morbidity compared to open repair for the treatment of rTEF and it can be used as a first line option with the disadvantage of repeated treatment attempts to achieve complete closure of the fistulas. In our case, surgical management was mandatory because of the previous failure to close the fistula with less invasive techniques (clips application) and due to the persistent air leak during mechanical ventilation that resulted in patient deterioration.

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### **Citation of this Article**

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