#### **CASE REPORT**



# Acquired tracheo-esophageal fistula in adult—a classical case of 'what not to do'

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### **Abstract**

Acquired tracheo-esophageal fistulas (TEFs) are challenging. The most common causes are prolonged intubation, malignancy, and trauma whereas granulomatous infections like tuberculosis are rare. Endoscopic intervention with esophageal or tracheal stenting or clipping is of unproven benefit in the management of such lesions, where surgical repair is almost invariably required. We report a case of a 32-year-old man, with a case of multidrug-resistant pulmonary tuberculosis. He had no history of malignancy or trauma. The patient developed spontaneous TEF probably due to mediastinal lymph node necrosis. Multiple attempts were made using staplers, clips, and atrial septal defect (ASD) device closure but were unsuccessful. The nuanced complication leads to very individualized course of treatment which was optimal for this patient.

**Keywords** Tracheo-esophageal fistula · Mediastinal lymph node · Tuberculosis

# Introduction

Acquired tracheo-esophageal fistula (TEF) is defined as a pathological connection between the trachea and the esophagus that results from a spectrum of disease processes. The major causes of acquired non-malignant TEFs include postintubation injury, chronic infections (e.g., tuberculosis), radiation injury, and post-surgical lesions [1]. Several modalities like fibrin glue injection, atrial closure devices, and endoscopic repairs have been described in the management of TEF [2, 3]. Non-surgical interventions have very limited role in such lesions and lead to recurrence as seen in this case. Hence, early and definite surgical repair is almost invariably required.

# **Case report**

A 32-year-old male patient had come to us with complaints of dysphagia and coughing for 5 months. He has a known case of multidrug-resistant tuberculosis involving lung parenchyma

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and mediastinal lymph nodes 2 years back, for which he was treated and became asymptomatic. He had no history of trauma, mechanical ventilation, or malignancy; he was coronavirus positive and recovered well. On examination, there were bilateral basal crepitations. His initial blood investigations showed no abnormality. His sputum was negative for tubercle bacilli. Upper gastrointestinal endoscopy and bronchoscopy were done which showed TEF of 7-9 mm with opening of 2 cm above the carina at the tracheal end and at 25 cm from the incisors at the esophageal end. Computed tomography scan showed fistulous communication between the posterior aspect of the trachea and the anterolateral aspect of the esophagus measuring  $7 \times 5.4 \times 7$  mm post Koch's sequel with hilar and mediastinal lymphadenopathy. Initial attempts were made to repair TEF with clips and cardio-occlusive device. However, both of them were explanted during the episode of repeated coughing. Repeat gastrointestinal endoscopy was performed after previous failed attempts and defect size was found to be 10-11 mm, so decision for surgical correction was taken. Repair was performed via right posterolateral thoracotomy through the 4th intercostal space. Retropleural exposure was obtained and intercostal muscle flap was prepared. During the dissection, azygous vein and vagus nerve were identified and azygous vein was divided. There was extensive fibrosis and adhesions at the fistula site, so fistulous track was dissected and divided. The esophagus was identified and dissected, tracheal opening was also identified (Fig. 1), margins were

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freshened, and the esophagus was sutured using polydioxanone (4–0) in an interrupted manner and the trachea was sutured using polyglactin (3–0) in a horizontal mattress manner (Fig. 2). Post repair, there was no leak on performing saline test. Tracheo-esophageal groove was covered using intercostal muscle flap and fixed using polyglactin (3–0) (Fig. 3); pleural drain was kept. The patient's postoperative period was uneventful and he was discharged successfully. As per our institutional protocols, tube feeding was started on 5th postoperative day. The patient was started on oral feeding on the 7th day, but since oral intake was less, he was kept on a liquid diet and gradually upgraded to full feeds at the 3rd week. Lymph node biopsy from the pretracheal region was suggestive of chronic inflammation and showed no evidence of active tuberculosis or malignancy.

# Discussion

Acquired TEF is defined as a pathological connection between the trachea and the esophagus that results from a spectrum of disease processes. The TEF can be caused by either malignancy or benign conditions. TEF from malignancy accounts for more than 50% of acquired TEFs [1]. The major causes of acquired non-malignant TEFs include postintubation injury, chronic infections (e.g., tuberculosis), radiation injury, and post-surgical lesions [2, 3]. The management strategy for TEF should encompass multiple facets of the disease, which include identifying the underlying etiology, goals of care, and assessing the stability. This was carried out using imaging and endoscopic modalities to conduct preoperative assessment and ultimately choosing an endoscopic versus a definitive surgical therapy. Prior to undertaking interventions, it is crucial to determine and treat the underlying condition implicated in TEF formation. Several modalities

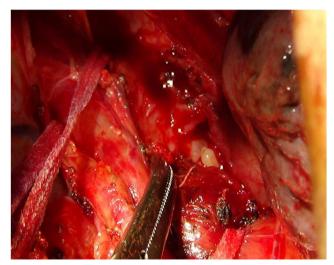


Fig. 1 Tracheo-esophageal fistula was dissected and opened tracheal end and esophageal end are seen

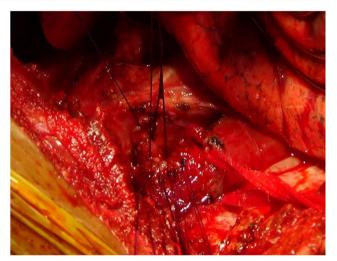


Fig. 2 The trachea and the esophagus were repaired

have been described in the management of TEF. These modalities are fibrin glue injection, atrial closure devices, and endoscopic repairs. Fibrin glue injections have been used to treat small fistula (< 5 mm). This method has high failure rate in larger fistula (> 8 mm) due to the rapid dissolution of the coagulative effect, leading to a recanalisation of the fistula. The atrial closure device, which was originally designed for trans-catheter closure of cardiac defects, has been used to treat TEFs, but significant airway complications have been reported with device usage, ranging from airway obstruction from mucostasis and granulation tissue formation with formation of new TEF from erosive changes relating to the device itself [4]. Buskens et al. experienced failure in endoscopic treatment when attempting with a fibrin glue or hemostatic clips even in a patient with a long mediastinal fistula track [5]. Several different surgical options are described for repair of a TEF, including direct closure of the tracheal and esophageal defects with or without a muscle flap, tracheal resection and

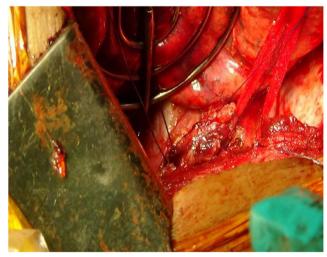


Fig. 3 Intercostal muscle flap was placed between the tracheoesophageal groove and fixed

anastomosis with primary esophageal closure, tracheal closure with a synthetic patch, and esophageal diversion [6]. Although endoscopic treatment is an attractive technique due to its minimal intervention and low morbidity rates, when compared to surgical repair, it is only applicable in the case of small fistulas. Muniappan et al. advocated prompt surgical repair to control the fistula, rather than depending on stents, because stents may lead to significant granulation tissue and typically extend the airway injury [7].

# **Conclusion**

Acquired TEF secondary to tuberculosis is insidious and needs thorough diagnosis. Early and definite surgical intervention is proposed once the diagnosis is confirmed. Non-surgical interventions have very limited role and leads to recurrence as seen in this case. Hence, surgical ligation of TEF is the most definitive form of management for benign conditions.

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#### **Declarations**

**Human and animal rights statement** All procedures performed in this study involving human participants were in accordance with the ethical standards of the institutional research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards. This article does not contain any studies with animals performed by any of the authors.

Ethics approval Not applicable.

**Consent to participate** Valid consent taken.

**Consent for publication** Proper consent taken.

**Conflict of interest** No potential conflict of interest relevant to this article was reported.

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