

# Tracheoesophageal Fistula Secondary to Tuberculosis: A Case Report

## Tüberküloza Sekonder Trakeoözefageal Fistül: Olgu Sunumu

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### Abstract

A tracheoesophageal fistula (TEF) is a pathological connection between the trachea and esophagus. Infectious diseases rarely lead to TEF, and tuberculosis is the most common cause of TEF among all infectious causes. A 78-year-old male patient under examination for esophageal malignancy at the gastroenterology service, and who was expected to undergo an endoscopic biopsy, was diagnosed with aspiration pneumonia after complaints of dyspnea and coughing after liquid intake, and was transferred to chest diseases clinic. TEF was identified from a bronchoscopy. Mycobacterium tuberculosis DNA was isolated during a bronchial aspiration. An endoscopic biopsy, performed after esophageal malignancy was suspected, gave the result of a granulomatous reaction. The patient was diagnosed with tuberculosis-induced TEF. Anti-tuberculosis treatment was initiated, and a tracheal Y stent was fitted. In this article we present a rare case in which a TEF emerged secondary to tuberculosis, and suggest that tuberculosis may be a cause of TEF. It is advised that before starting invasive surgical TEF treatment, it is essential to take the necessary precautions.

**Key words:** Aspiration pneumonia, Trachea esophageal fistula, tuberculosis, tracheal stent.

### Özet

Trakeaözofageal fistül (TÖF) trakea ile özefagus arasında patolojik bir bağlantı olmasıdır. Enfeksiyon hastalıklarına sekonder gelişen TÖF çok nadir nedenlerdendir. Tüberküloz enfeksiyonu ise enfeksiyöz nedenler arasında en sık izlenenidir. Gastroenteroloji servisinde özefagus malignitesi ön tanısı ile tetkik edilen ve endoskopik biyopsi sonucu beklenen 78 yaşında erkek hasta nefes darlığı, sıvı gıda aldıktan sonra olan öksürük yakınması olması üzerine yapılan tetkiklerde aspirasyon pnömonisi tanısı aldı ve göğüs hastalıkları servisine nakil alındı. Bronkoskopisi yapıldı. TÖF ağız görüldü. Bronş aspirasyonunda mycobacterium tuberculosis DNA'sı saptandı. Endoskopik biyopsi sonucu granülatöz reaksiyon olarak geldi. Hastaya tüberküloz nedeni TÖF tanısı konuldu. Anti-tüberküloz tedavisi başlanan hastaya trakeal Y stent takıldı. Makalemizi tüberkülozun nadir görülen bir TÖF nedeni olması ve nedeni bilinmeyen TÖF'ün operasyonu öncesinde tüberküloz bulaşına yönelik gerekli önlemlerin alınmasının önemini vurgulamak amacıyla sunuyoruz.

**Anahtar Sözcükler:** Aspirasyon pnömonisi, trakeoözefageal fistül, tüberküloz, trakeal stent.

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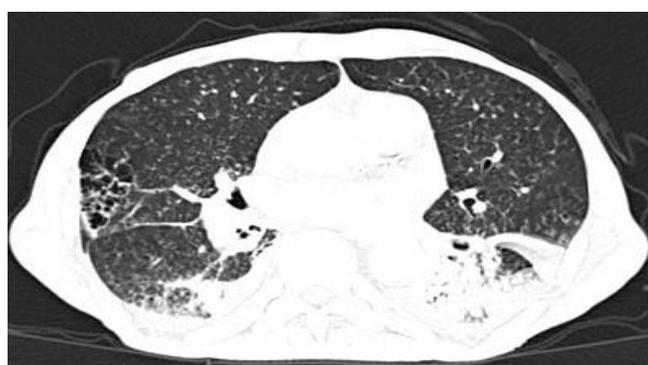
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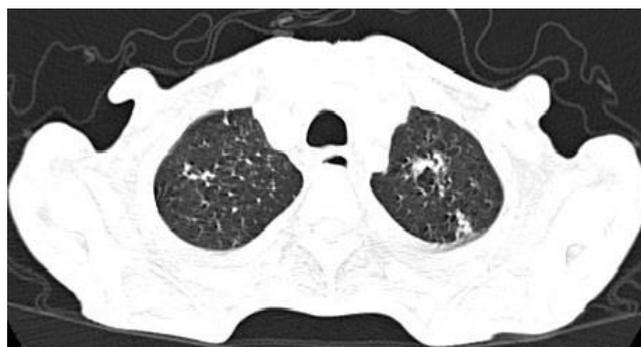
A tracheoesophageal fistula (TEF) is a rare pathological tract between the trachea and the esophagus. TEF can be either congenital or acquired, although the former is more common, occurring as a result of an abnormality in the development of the septum between the ventral tube and the dorsal esophagus in the embryological period, and is observed in around one of every 5,500–6,000 births (1,2). Acquired TEF can develop secondary to tracheal intubation, malignancy, granulomatous mediastinal infections, immunodeficiency syndromes, open and closed chest traumas, and iatrogenic trauma. Primary esophagus or lung cancers, or the treatment of such mediastinal tumors as lymphoma, account for 50–70% of all acquired TEF cases, while TEF development due to tuberculosis is a rare condition. The most significant symptom is recurrent coughing followed by fluid intake. Aspiration pneumonia, sepsis and mediastinitis secondary to recurrent aspirations are the most severe complications, causing severe morbidity and mortality. Emergency treatment should be planned as soon as TEF is diagnosed. In this article we present a case study and the treatment plan of a patient who was diagnosed with TEF secondary to tuberculosis, which is very rare.

## CASE

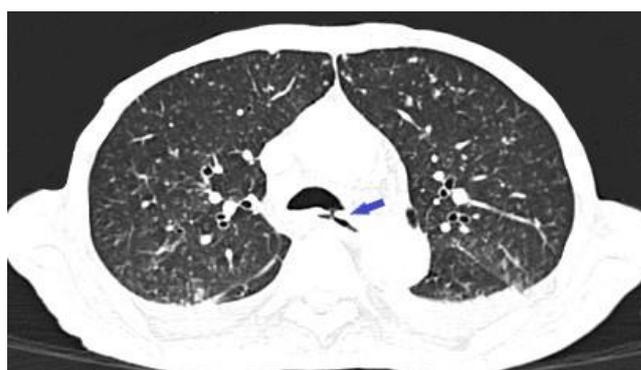
A 78-year-old male patient was admitted to the gastroenterology department with a pre-diagnosis of esophageal carcinoma. During hospitalization, the patient revealed the following symptoms: dyspnea, cough, difficulty in swallowing solid-liquid foods, and sputum. He was transferred to the chest diseases clinic with a preliminary diagnosis of aspiration pneumonia. The patient's history and family history were normal. Upon physical examination, the patient was dehydrated and cachectic, and the skin turgor tone was decreased.



**Figure 1a:** Consolidated areas including air bronchograms in the lower lobes of both lungs but more dominant on the left side



**Figure 1b:** Nodular appearance in the upper lobes of both lungs



**Figure 1c:** A connective structure between the left main bronchus posterior and esophagus suggesting TEF

A respiratory system examination revealed the presence of rales in the lower areas of both lungs. Laboratory tests revealed the following results: Hb: 10.1 g/dl, platelet: 109,000 dL, urea: 69 mg/dl, AST: 42 U/L, ALT: 52, Ca: 7.6 mg/dl, protein: 4.4 g/dl, albumin: 2.1. CRP value was increased (20.92 mg/dl), other parameters were in the normal range.

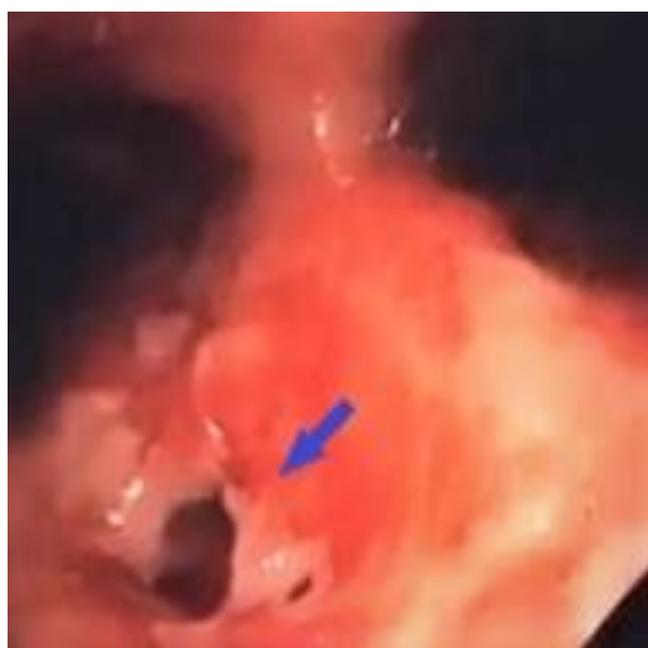
A scattered patchy opacity was observed in both lungs in a postero-anterior chest X-ray. Ceftriaxone 1x2 gr, clindamycin 600 mg 3x1 IV and total parenteral nutrition treatments were initiated. An endoscopy revealed a mucosal ulceration at the 25th cm of the esophagus and a 9 mm scope was unable to pass through this narrowed passage. A thorax computed tomography (CT) of the patient revealed consolidated areas that included air bronchograms in the lower lobes of both lungs that were more dominant on the left side (Figure 1a), as well as a peribronchial nodular appearance in the upper lobes of both lungs (Figure 1b). In the mediastinal frame, retrotracheal, left lower paratracheal and subcarinal lymph nodes were identified. Esophageal wall thickness was also increased, and a fistula was identified between the posterior tracheal wall and the esophagus suggesting, TEF at the carinal level (Figure 1c).

During the performed bronchoscopy, two large fistula orifices were observed at the posterior of the left main

bronchus (Figure 2a and b). Dense mucopurulent sputum was aspirated, especially from the left main bronchus inlet. In a bronchial aspiration, an acid-resistant bacillus (ARB) was negative, but *Mycobacterium tuberculosis* DNA was detected via a polymerase chain reaction (PCR). An NG catheter was inserted to continue feeding. The patient consulted a thoracic surgeon, and after further evaluation, the patient was considered suitable for a tracheal stent application. A tracheal Y stent was subsequently applied via rigid bronchoscopy.



**Figure 2a:** Two fistula orifices were observed at the posterior of the left main bronchus



**Figure 2b:** Two fistula orifices were observed at the posterior of the left main bronchus

Endoscopic biopsy specimens showed excessive fibrinopurulent exudate as well as ulceration, a squamous epithelial fragment with reactive cellular changes, and a granulomatous reaction. No neoplastic epithelial cell infiltration was detected. The patient was diagnosed with TEF secondary to tuberculosis. Anti-tuberculosis treatment was initiated, and the patient was transferred to the tuberculosis clinic.

Endoscopy was performed again for further investigation. An approximate 7–8 mm TEF orifice was observed 25–26 cm distant to the cutting teeth. NG provided nutrition and PEG was planned for long-term feeding. The patient was consulted by a gastroenterologist. The patient was referred to the center where the PEG procedure was performed. After not attending the follow-up, the patient was reported to have been found dead at home after a month.

## DISCUSSION

Both the diagnosis and treatment of TEF are difficult for clinicians. TEF frequently presents with recurrent lower respiratory tract infections. The most common symptom is recurrent coughing, especially after fluid intake. Some patients may suppress coughing by lying in the supine position (Ono sign). Digestive system symptoms are rarer, with epigastric sensitivity and reflux being the most common, resulting from the stomach filling with air during expiration (3,4). Recurrent aspirations and aspiration pneumonia may also lead to such serious complications as sepsis and mediastinitis. For this reason, oral intake should be ceased immediately if TEF is suspected. Our case presented with a recurrent cough and aspiration pneumonia after fluid intake; and oral intake was stopped immediately.

Percutaneous vertebroplasty has been widely used for the operative treatment of vertebral fractures over the past 30 years, having gained popularity as a method bringing immediate pain relief (6,7). Transvertebral cement leakages into the surrounding tissues and into the paravertebral veins are common complications after percutaneous TEF can be classified as congenital or acquired. Some 49% of the acquired causes are malignancies, while the remaining are trauma, tuberculosis, actinomycosis and esophageal diverticulum (5). Tuberculosis-induced TEF cases are rare (6), while tuberculosis-dependent TEF may occur due to endobronchial tuberculosis, mediastinal lymph node tuberculosis or esophageal tuberculosis.

Tuberculoid mediastinal lymphadenopathy, which can cause necrosis and perforation of the adjacent structure, is an important pathogenetic factor in fistula formation (7). Esophagus-originated tuberculosis is usually seen in immunocompromised patients, and may cause fistulas by abrading the adjacent tracheobronchial tree (8). Although our patient presented with radiologic parenchymal tuberculosis finding, no pathological area was found that suggested endobronchial tuberculosis in bronchoscopy. The most probable hypothesis for our patient is that an infection in the mediastinal pathological lymph nodes between the esophagus and the trachea created a fistula between the trachea and the esophagus, although another possibility is that it was induced by endobronchial tuberculosis due to the weakening of the immune system of this old patient.

The opening of the fistula is usually located on the posterior wall of the trachea. In our patient, two large openings, one bigger and one smaller, were observed at the posterior of the left main bronchus entrance at the carina level. Esophageal tuberculosis is mostly associated with mediastinal lymphadenopathy, and tracheo-oesophageal and/or esophageal-mediastinal fistulas are present in half of such cases (9). Our patient was initially thought to have an esophageal carcinoma, but was eventually diagnosed with a TEF. Although esophageal malignancy was thought to be the cause, the endoscopic biopsy resulted with a granulomatous reaction, and so it was considered that the cause of the TEF may be a non-malignant, granulomatous infection, such as tuberculosis. The detection of the DNA of Mycobacterium Tuberculosis in the deep tracheal aspiration via PCR confirmed the diagnosis of tuberculosis.

Although surgery was the most common treatment in the past, endoscopic and bronchoscopic stent applications are today the most common modalities. The risk and difficulty of surgery and the long anesthesia times play a key role in surgical decisions as a second option (10). Laser and argon plasma coagulation are other options for the treatment of small fistulas, but due to the potential for the expansion of the fistula, they should be applied by experts, taking due care (11). Treatment should be planned as soon as possible after the confirmation of a TEF diagnosis, and the primary cause should be ascertained, after which, the treatment of the primary cause should be added to the interventional therapies. This approach decreases both mortality and morbidity rates. In our patient, after confirming the diagnosis of TEF, intervention modalities were performed immediately to prevent

aspiration. After the primary cause was found to be tuberculosis, anti-tuberculosis treatment was started.

In countries such as ours, where the incidence of tuberculosis is still high, tuberculosis should be considered in the etiology of TEF. Owing to the need for an interventional procedure for the immediate repair of a fistula, and the amount of time needed for a definitive diagnosis of tuberculosis being long in patients with TEF, it is essential that health workers remember that tuberculosis is a cause of TEF, and to take the necessary precautions prior to surgical and bronchoscopic procedures.

## CONFLICTS OF INTEREST

None declared.

## AUTHOR CONTRIBUTIONS

Concept - M.O.G., M.G., N.A., K.C.C., D.A.; Planning and Design - M.O.G., M.G., N.A., K.C.C., D.A.; Supervision - M.O.G., M.G., N.A., K.C.C., D.A.; Funding - M.G., N.A., M.O.G.; Materials - M.O.G., M.G., N.A.; Data Collection and/or Processing - M.O.G., M.G.; Analysis and/or Interpretation - M.O.G., M.G.; Literature Review - M.O.G., M.G.; Writing - M.O.G.; Critical Review - M.O.G., M.G.

## YAZAR KATKILARI

Fikir - M.O.G., M.G., N.A., K.C.C., D.A.; Tasarım ve Dizayn - M.O.G., M.G., N.A., K.C.C., D.A.; Denetleme - M.O.G., M.G., N.A., K.C.C., D.A.; Kaynaklar - M.G., N.A., M.O.G.; Malzemeler - M.O.G., M.G., N.A.; Veri Toplama ve/veya İşleme - M.O.G., M.G.; Analiz ve/veya Yorum - M.O.G., M.G.; Literatür Taraması - M.O.G., M.G.; Yazıyı Yazan - M.O.G.; Eleştirel İnceleme - M.O.G., M.G.

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