

# Esophageal-Pleural Fistula: the Cause or Effect of Recurrent Pneumonia?



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## ABSTRACT

Esophageal-pleural (E-P) fistula is introduced to be an uncommon clinical finding maybe due to non-specific clinical pointers and vague radiologic clues. In this regard, the main drivers are esophageal traumatization, endoscopic dilation, irradiation and corrosive agent ingestion as well as major underlying esophageal diseases like carcinomas and tuberculosis. The presented case is a 51-year-old man with medical record of recurrent pneumonias resulted in right lobectomy who admitted to the medical institution with a significant empyema leading to chest tube insertion. It should be noted that the presence of food particles in the chest bottle in addition to the history of previous surgery raised suspicion of a post-surgical E-P fistula formation. Attempts were made to find fistula tract and insert an esophageal stent bypassing the fistula opening. However, investigations argued against E-P fistula as an ensuing event after surgery and arose the question that whether E-P fistula would be the cause or effect of pneumonia in the presented case?

## Introduction

**A**fter pneumonectomy, esophageal traumatization, tuberculosis and other inflammatory processes, E-P fistula has rarely been reported. [1] Diagnosis of E-P fistula has always been challengeable due to difficult distinction on medical imaging and indistinguishable clinical characteristics. A true medical history would help

more specific investigations for detection of such fistulas. The history of lobectomy due to unresolved recurrent pneumonias in addition to the presence of food debris in the recently inserted chest tube for severe empyema raised suspicion of E-P fistula in the presented patient. Endoscopic investigation was made in an effort to find E-P fistula and insert an esophageal stent. Surprisingly, careful evaluations gave rise to another reason for E-P fistula instead of surgical manipulation of pleura which could also

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justify patient's symptoms before surgery.

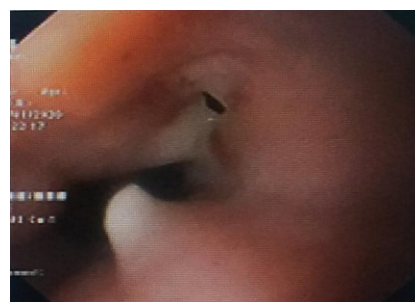
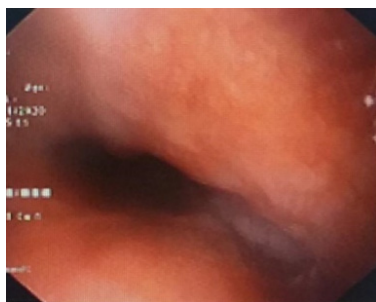
### Case presentation

A 51-year-old man with history of smoking 20 pack/year presented with a long-term history of severe cough, sputum, tachypnea, right-sided chest pain, fatigue and weight loss which was aggravated 3 days before admission. He denied any history of alcohol or other illicit drugs. He has had a history of recurrent bouts of severe pneumonias for years which finally resulted in right upper lobectomy about 4 years before recent admission. His recurrent pneumonias had been attributed to the complicated right upper lobe abscess formation unresponsive to broad spectrum antibiotics and long-term chest tube insertion. Therefore, it was decided to proceed with a right upper lobectomy. Specimens were analyzed which were negative for malignancy and tuberculosis. Complementary revisions were negative for vasculitis, neoplastic lesions, any suspicious drug history and hypersensitivity pneumonitis. Total IgG, IgA, IgM levels were also within normal limits. Since then, he continued to deteriorate and were frequently managed for post-lobectomy empyema. On admission, he spiked a temperature of 39°C and initial chest X-ray and CT scan revealed right hydropneumothorax with atelectasis of the right middle lobe and empyema for which he was treated with empiric wide-spectrum intravenous antibiotics, inhaled bronchodilators and intercostal tube insertion. Physical examination was notable for right decreased sound due to previous lobectomy and hydropneumothorax. On lab tests, leukocytosis and elevated ESR & CRP were remarkable. Bacteriology was positive for klebsiella pneumoniae and cytology was negative for malignant cells. 4 days after recent admission, food debris was noted in his chest bottle which made us proceed with barium esophagography looking for an esophageal-pleural fistula which proved the diagnosis. Given the previous history of right upper lobectomy, fistula was attributed to the surgery. The patient was started on total parenteral nutrition in addition to antibiotic therapy. Attempts

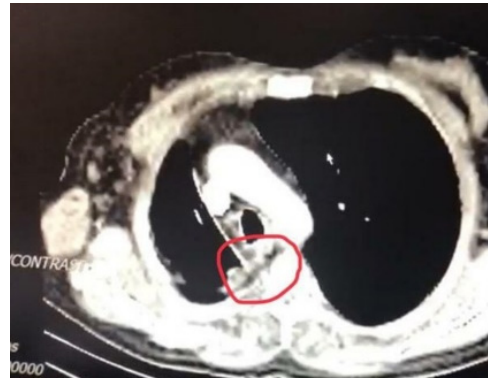
were made to find fistulous tract via endoscopy and implement an esophageal stent bypassing the fistula. The scope was inserted into the oropharynx down to the esophagus which revealed a diverticulum at the proximal part of esophagus (20cm from incisors) and fistula opening just distal to the diverticulum (25cm from incisors) (Fig. 1A, B). It had appeared to be a bony lesion exactly facing the fistulous tract. Taken together, it was felt that an embedded bone in the esophageal wall (maybe years before onset of symptoms in the form of pneumonia) would be the cause of E-P fistula and recurrent pneumonia in the presented case.

### Discussion

E-P fistula after pneumonectomy for malignant diseases, tuberculosis and other inflammatory processes has rarely been reported. [1] Per the revised studies, precise diagnosis of E-P fistula has been made utilizing radiographic contrast media or methylene blue. Observation of food particles in the pleural space and chest tube has also been reported to be diagnostic. [1] The latter was detected in the presented case which raised suspicion of an E-P fistula tract. It was then confirmed by barium study. Pleural space manipulation during surgery is one of the most known etiologies for E-P fistula. It is reported that left-sided pneumonectomies harbor lower probability of fistula formation than right-sided ones. Duration of E-P fistula formation after pneumonectomies has been reported to be early up to 25 years. [2,3] Early formation of fistulous tract is generally been attributed to the surgical errors and the reasons for late-onset E-P fistulas are still in question. [4, 5] Hence, various durations of fistula formation reported in the literature after surgery indicate that time suffers from poor sensitivity as a diagnostic tool to differentiate E-P fistula from other causes of recurrent pneumonia in the presented case. Spontaneous rupture of esophagus (Boerhaave's syndrome) has also been introduced as a rare cause of E-P fistula formation. [6] In contrast to the popular belief in the presented case that E-P



**Fig. 1A.** A small diverticulum was detected just above the fistula in the upper esophagus. **B.** Fistula tract opening was found in the upper esophagus facing an embedded bony lesion.



**Fig. 2.** Small amount of contrast media extravasation from esophagus into the pleural space is noted.

fistula might be resulted from lobectomy, evidences suggested another reason for fistula formation and raised suspicion of presence of fistulous tract before lobectomy. Endoscopic evaluation revealed the presence of an embedded bony lesion in front of the opening of fistulous tract (Fig. 1A, B). Now, the question is whether an embedded foreign body could stay for a long time without mediastinitis and other signs and symptoms of esophageal perforation or not. Despite the fact that esophagus was perforated freely into the pleural space, no evidence of acute mediastinitis was seen in the presented case. This was comparable with introduced cases by Richardson, Campbell and Trinkle. [7] This strange clinical response is explainable with slow progression of fistulous tract formation over the years making pleural space be exposed to the gastrointestinal tract contents slowly and steadily. Thus, mediastinal drainage was not necessary in the mentioned patient up to the recent admission. Foreign bodies penetrated and embedded in the gastric wall are reported more commonly than esophagus wall. Similar to the case report of Guo-Dong Shan et al. who reported granuloma formation due to an embedded foreign body in the stomach mimicking a submucosal tumor. [8] However, there are few reports on embedded foreign bodies in the esophagus without mediastinitis and warning signs & symptoms of perforation slowly progressing into the fistulous tract. [9] Actually, the relationship between esophagus and pleura is related to the amount of mediastinal fat and connective tissue which are critical determinants of patient's symptoms specifically in a neglected foreign body ingestion. [10] These instances show that a foreign body would pass through the mucosal surface and stimulate re-epithelialization of surrounded mucosa if it did not pass through the gastrointestinal tract or did not perforate the esophagus. [11] Diagnosis needs a precise history-taking and non-contrast imaging. The true diagnosis (the observation of foreign body in the esophagus) in the presented case had been obscured by non-specific symptoms which were amenable to

only contrast-enhanced CT scans for evaluation of underlying cause of pneumonia. This emphasizes the need for a non-contrast imaging in patients with recurrent pneumonias even without a clear history for foreign body ingestion. Revisions of previous contrast-enhanced studies before surgery revealed small amount of extravasation of contrast media into the pleural space in limited cuts which was neglected due to inexistence of prodromal manifestations of perforation in the patient (Fig. 2). Three-dimensional studies would help a better localization of detected foreign body as well as contrast-enhanced studies which precisely evaluate E-P fistula. [12]

Likely etiologies for recurrent pneumonias are anatomical anomalies, underlying inflammatory or infectious parenchymal diseases, congenital or acquired immunodeficiencies which all were negative in the presented case. So, further evaluation to find the reason for recurrent pneumonia in right upper lobe was required. [13] The location of repeated pneumonias is also suggestive of underlying etiology. Foreign body inhalation, tumoral growth, and pulmonary sequestration are related to one anatomical involvement in the lung. Large distinct involvement of parenchyma is attributed to the chronic obstructive pulmonary disease, bronchiectasis and asthma. Recurrent infections in different locations are seen in primary or secondary immunodeficiencies like common variable immunodeficiency and acquired immunodeficiency syndrome. Aspiration pneumonia should also be taken into account for patients with recurrent pneumonias in basal segments of lower lobes and posterior portions of upper lobes. [14] Accordingly, the location of infection in the presented case reinforced the diagnosis of an anatomical aberrancy like E-P fistula as what we found in the later investigations. Large abscess formation unresponsive to treatment on that time frame finally resulted in lobectomy in the presented case. [15] Nevertheless,

investigations should have been continued up to detection of a logical etiology justifying non-resolving abscess formation and recurrent pneumonias in one lobe. This is the missed point in the presented case. Due to the presence of an embedded foreign body in the esophagus wall as well as a large fistulous tract opening, surgery was considered for the patient and esophageal stent was found to be useless and was not inserted for the patient. Surgical consultation ended up esophagectomy and gastric pull-up. After that, he appeared to feel better and stayed asymptomatic up to the next visit.

## Conclusion

The lesson learned from this manuscript is that foreign body ingestion and ensuing fistulous tract formation should be taken into account in the case of recurrent pneumonias even without typical symptoms of perforation. Thus, chest CT scan with and without contrast enhancement is recommended for these patients.

## Ethical Considerations

### Compliance with ethical guidelines

There were no ethical considerations to be considered in this article.

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### Conflict of Interests

The authors have no conflict of interest to declare.

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