Unusual Presentation of an Esophagopleural Fistula in a Patient With Severe Esophagitis: Blunt Trauma or Iatrogenic?

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ABSTRACT
Esophagopleural fi stula (EPF) is rare despite the anatomical proximity of the esophagus and the pleural space. A 64-year-old man presented with a pneumothorax after a fall requiring chest tube placement. An esophagastroduodenoscopy revealed severe LA grade D esophagitis and a large EPF in the distal esophagus. Computed tomography scan revealed that subtle tracking of air extending from the distal esophagus into the right pleural space was noted. The patient was treated with placement of a fully covered esophageal metal stent, and he recovered uneventfully. Interpreting key subtle clues in pleural fluid analysis and imaging can lead to a timely diagnosis and thus improves morbidity and mortality of EPF.

INTRODUCTION
Esophagopleural fistula (EPF) is a rare entity. It refers to an abnormal connection between the esophagus and the pleural space. It is an uncommon condition despite the anatomical proximity of these structures. Although rare, it is associated with high morbidity and mortality if not diagnosed promptly. Diagnosis can be challenging and can be missed in regular computed tomography (CT) images. We present a case of EPF with an atypical presentation and demonstrate how a high index of suspicion and interpretation of key subtle clues in imaging and laboratory tests lead to a prompt diagnosis.

CASE REPORT
A 64-year-old man with a history of alcohol abuse presented to our trauma center after falling from his wheelchair down several stairs 19 hours before. He was transferred from a hospital where a chest tube was placed after he was found to have a large pneumothorax. On arrival, the patient was alert and fully oriented, complaining of right-sided chest pain. He was afebrile, tachycardic, and tachypneic, requiring 4 L of oxygen using a nasal cannula. Physical examination was significant for decreased breath sounds over the right lower lobe and left-sided hemiplegia from a previous stroke. The chest tube drainage was initially described as serosanguineous as per outside records.

A CT scan was obtained upon admission. Laboratory tests showed no leukocytosis, and pleural fluid analysis revealed red blood cell 350 cells/μL, white blood cells (WBC) 1,750 cells/μL, protein 0.9 g/dL, glucose 1,088 mg/dL, lactate dehydrogenase (LDH) >1,300U/L, pH 5.56, and adenosine deaminase (ADA) 10.6 U/L. One of 2 pleural fluid cultures was positive for Rothia mucilaginosa, Granulicatella adiacens, and Candida albicans that were thought to be contaminants. For a week, the patient continued to be afebrile, but the chest tube was noted to have a high output of 2-4 L per day of grossly milky-appearing fluid. He was started on broad-spectrum antibiotics. Because of the fluid appearance and persistent high output through the chest tube, the possibility of a thoracic duct injury was raised. The pleural fluid was sent for triglyceride and chylomicron levels. Lymphangiography with interventional radiology embolization was planned while awaiting the procedure, and on day 10 of admission, the patient had an

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episode of melena associated with a drop in hemoglobin from 10 to 4.5 g/dL. He was transferred to the intensive care unit because of hemorrhagic shock and subsequently had cardiac arrest with successful return of spontaneous circulation after 5 minutes. He was intubated during the cardiac arrest. Intravenous fluids and 4 units of packed red blood cells were given. Vasopressor support and intravenous proton pump inhibitor drip were started.

The gastroenterology service was consulted for an esophagogastroduodenoscopy (EGD), which was performed the same day. It revealed severe esophagitis from the mid to the distal esophagus along with a large fistulous tract opening extending from the distal esophagus to the right pleural space, and on advancing an adult gastroscope through this fistulous tract, the end of the chest tube along with a few old retained pills were seen (Figure 1). The area surrounding the fistulous tract was biopsied, and the specimens were sent for histopathology and for acid-fast bacilli smear and fungal and viral culture. The biopsies were negative for malignancy, and the cultures were all negative. After the EGD, the CT scan obtained on admission was retrospectively reviewed, looking for evidence of EPF. Soft tissue window and axial lung window images of the chest revealed tenting of the distal esophagus and subtle tracking of air extending from the distal esophagus into the right pleural space, confirming the presence of EPF (Figure 2).

A repeat EGD was performed the following day with the placement of a 20-mm-wide 10-cm-long fully covered metal esophageal stent (Figure 3). The proximal end of the stent was secured to the esophageal mucosa with 5 endoclips to decrease the risk of stent migration. One day after stent placement, the patient underwent right-sided video-assisted thoracoscopic surgery, drainage of pleural effusion, pleural debridement, and decortication. He was subsequently weaned off the vasopressors and successfully extubated. Antibiotics and antifungals were given for 4 weeks after video-assisted thoracoscopic surgery, as recommended by the infectious disease service. The patient was kept nothing by mouth. A jejunostomy tube was placed by interventional radiology for long-term enteral feeding while allowing the fistulous tract to heal. The jejunostomy tube was chosen over a gastrostomy tube because of the findings of severe reflux esophagitis. The patient was then discharged to a long-term acute care facility. An esophagogram performed 2 months after stent placement showed closure of the fistulous tract with no extravasation of contrast. The chest tube was removed. EGD was then repeated under fluoroscopy, and the esophageal stent was removed. Contrast injected through the scope into the esophagus confirmed complete closure of the fistulous tract opening (Figure 4). An ulcer was seen at the previous site of the fistulous tract. A high-dose proton pump inhibitor was continued. The patient recovered uneventfully.

**DISCUSSION**

EPF refers to an abnormal connection between the esophagus and pleura. It is an uncommon condition despite the anatomical proximity of these structures. The most common etiologies include malignancy of the esophagus, lung, or mediastinum. It is caused either by direct tumor invasion or by subsequent perforation after radiation, laser therapy, chemotherapy, or pre-existing stents. Cases of EPF were also reported after pneumonectomy, probably related to intraoperative esophageal injury and wall thinning and iatrogenic trauma related to esophageal instrumentation or esophageal wall ballooning. Benign EPF is rare and mainly caused by trauma or infection. The presentation could mimic either a primary respiratory disorder or a gastrointestinal disorder. Patients can present with fever, retrosternal chest pain, dyspnea, dysphagia or nausea, and vomiting. We report a rare case of EPF in a patient with severe esophagitis. The exact etiology of the formation of EPF in our patient remains uncertain.

One hypothesis is that the initial insertion of chest tube iatrogenically caused damage of the parietal pleura and penetrated the esophageal wall. This hypothesis remains difficult to prove because of the initial presentation of tension pneumothorax and the emergency circumstances, and no CT scan of the chest was obtained at an outside institution before chest tube placement. The other hypothesis is that the spontaneous rupture or tear of

![Figure 1](image1.png)  
**Figure 1.** Esophagogastroduodenoscopy showing (A) the gastroesophageal junction (white arrowhead) and the fistulous tract opening (white arrow), and (B) the endoscopic view of the end of the chest tube in the pleural space (black arrowhead) and a few old retained pills (black arrow).
the esophagus in the setting of severe esophagitis was possibly aggravated by retching and vomiting in this patient with alcohol abuse. There have been case reports of spontaneous transmural esophageal rupture in patients with severe esophagitis, thought to be related to the chronic inflammatory process, leading to the friability of the esophageal mucosa. However, in these cases, patients mostly had dysphagia, chest, or abdominal pain, none of which were present in our patient. Moreover, the presence of the fistulous tract on the initial CT scan performed before the EGD rules out the possibility that this was caused by esophageal instrumentation.

Diagnosis of EPF can be challenging, and it can be easily missed in regular CT scan images. Oral contrast given at the time of CT can increase the diagnostic yield when EPF is suspected. Other clues to the diagnosis are the presence of oral flora organisms in the pleural fluid culture. In our case, *R. mucilaginosa*, *G. adiacens*, and *C. albicans* grew in one of the fluid cultures. The pH of 5.56 in the pleural fluid was another clue to the diagnosis because a pleural fluid pH of 5–6 has been described in cases of esophageal rupture. A pH less than 7 is also associated with empyema and parapneumonic effusions, but in this condition, the glucose level is usually low. The pleural fluid glucose in our case was 1,088 mg/dL, most likely from contamination of the pleural fluid with enteric contents from the patient’s oral intake. The endoscopic treatment of EPF includes placement of a fully covered metal stent—like the one used in our patient. Over the scope clip placement has also been described with success. In conclusion, a high index of suspicion is necessary to diagnose this rare entity; interpreting key subtle clues in pleural fluid analysis and imaging can lead to a timely diagnosis.

**DISCLOSURES**

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