

## Case Report

# Spontaneous esophageal-pleural fistula

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## ABSTRACT

Spontaneous esophageal-pleural fistula (EPF) is a rare entity. We describe a case in a middle-aged female who presented with severe retrosternal chest pain and shortness of breath. Chest computed tomography showed right EPF and hydropneumothorax. She was managed conservatively keeping the chest tube drainage and performing feeding jejunostomy. A brief review of the imaging finding and management of EPF is discussed.

**KEY WORDS:** Esophageal injury, pleural fistula, esophagus perforation

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## INTRODUCTION

Esophageal-pleural fistula (EPF) is a rare entity which is formed secondary to esophageal instrumentation, surgery, malignancy, or as a rarer complication of postpneumectomy.<sup>[1-6]</sup> Spontaneous development of fistula between esophagus and pleura is very rare. Contrast-enhanced computed tomography (CT) is the very useful imaging technique to evaluate esophagopleural fistula.<sup>[6]</sup>

## CASE REPORT

A 40-year-old female presented with severe retrosternal chest pain with vomiting for four days. It was noncolicky, radiating to back, and there was no aggravating or relieving factors. The pain was associated with shortness of breath. There was no history of the upper gastrointestinal endoscopy or esophageal instrumentation. On chest examination, air entry and vocal resonance were decreased on right side. Chest radiograph showed moderate right pleural effusion. Chest tube was inserted which drained fluid containing ingested food particles. Chest CT [Figure 1] showed linear fistulous tract from right lateral aspect of

midthoracic esophagus to the right pleural cavity suggestive of EPF. There was no definite evidence of mediastinitis, or significant hilar or mediastinal lymphadenopathy. There was right hydropneumothorax with chest tube *in situ* and collapse-consolidation of the adjoining right lower lobe. In addition, there was volume loss on right hemithorax as compared to left side with associated pleural thickening. Esophagography [Figure 2] with nonionic contrast was done to demonstrate the exact site and size of fistula, showed approximately 2 cm right EPF at the level of junction of mid and lower thoracic esophagus. There was no evidence of contrast extravasation into mediastinum or left pleural cavity. Upper GI endoscopy was also done to rule out any associated esophageal inflammation or malignancy. It showed a fistulous opening at 27 cm from incisors with healthy surrounding mucosa. Hence, with the final diagnosis as spontaneous EPF, she was managed conservatively keeping the chest tube drainage and performing feeding jejunostomy. Definite surgery could not be performed as the patient lost to follow-up even after six months.

## DISCUSSION

Esophageal injury may be caused by iatrogenic trauma (esophageal instrumentation or external trauma), diseases of the esophagus such as corrosive esophagitis, esophageal ulcer and neoplasm, and rarely spontaneously.<sup>[1-6]</sup> Spontaneous rupture of esophagus or Boerhaave's syndrome usually results from sudden rise of intraesophageal pressure (due to contraction of cricopharyngeus muscle and closing of pyloric sphincter) associated with forceful vomiting or retching.<sup>[5]</sup> Mallory-Weiss tears are mucosal

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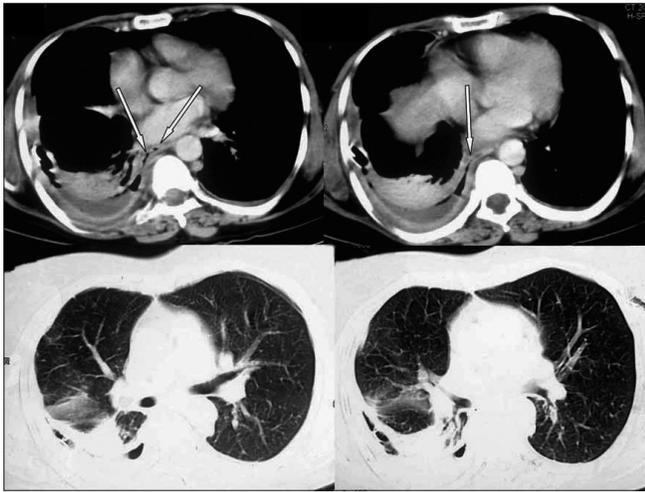


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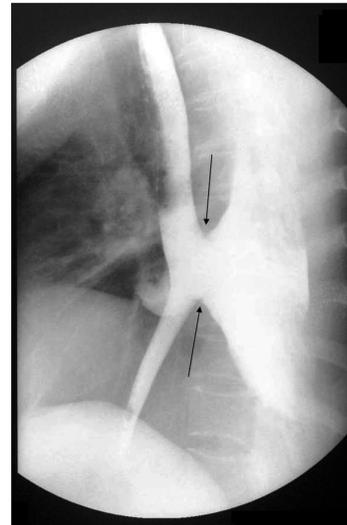
**Figure 1:** Axial sections of chest CT showing right hydropneumothorax with collapse-consolidation of the adjoining right lower lobe and chest tube *in situ*. Linear fistulous tract (arrow) is seen from mid-thoracic esophagus to the right pleural cavity, which is suggestive of esophageal-pleural fistula

tears caused by forceful or long-term vomiting, retching, or coughing. The patients present with gastrointestinal bleeding and these tears usually heal spontaneously.

EPF is an uncommon complication of iatrogenic trauma (endoscopic instrumentation) or postpneumonectomy.<sup>[1-6]</sup> The postpneumonectomy EPF are caused by surgical injury, local cancer recurrence, and chronic inflammation or infection.<sup>[4,6]</sup> The site of perforation depends on the cause. The possibility of direct EPF and extent of mediastinitis are determined by anatomic relationship of esophagus to the pleura, the amount of mediastinal fat and intervening connective tissue.<sup>[2]</sup> The diagnosis of EPF is difficult as the clinical sign and symptom are nonspecific. Esophagus injury should be considered when patient presents with retrosternal chest pain, fever, dysphasia, and dyspnea, especially when the patient gives antecedent history of esophageal instrumentation or surgery.

The radiological signs of the EPF depend upon site, duration, and severity of perforation; and more importantly, the integrity of pleura.<sup>[2]</sup> If the pleura remain intact, mediastinitis sets in with subsequent occurrence of rupture of mediastinal pleura, pneumothorax, or hydrothorax.<sup>[1,2]</sup> If the pleura is not intact, EPF can occur with resultant decompression of esophageal contents into the pleura and thus mediastinum may not be involved.<sup>[1]</sup> The diagnosis of esophageal rupture/EPF is made clinically; however, for confirmation, the imaging is required. The imaging modalities include chest radiograph, ultrasound, barium swallow, contrast-enhanced CT, and MRI with each modality having its advantages, and chest CT is very useful modality.

The findings on the chest radiograph are nonspecific and include pleural effusion, pneumothorax, or hydropneumothorax. Pneumomediastinum or



**Figure 2:** Esophagogram done with nonionic contrast showing approximately 2 cm right EPF (arrow) at level of junction of mid and lower thoracic esophagus. There was no evidence of contrast extravasation into mediastinum or left pleural cavity

subcutaneous emphysema is seen late in the course of the disease when pleural pressure exceeds mediastinal pressure. CT shows the exact extent of mediastinum involvement, confirms the X-ray findings, and can differentiate pleural from pulmonary disease. Other nonspecific CT findings include focal thickening or ballooning and thinning of the esophageal wall at the site of perforation.<sup>[2,3]</sup> Rarely small rent along the esophageal wall can be seen communicating with diseased pleural cavity. Contrast medium, if given orally, seen in the pleural space is pathognomonic sign of the entity. CT also helps in the management of the EPF as its prognosis depends upon the extent of the mediastinal involvement. EPF without mediastinal involvement usually carries good prognosis and may respond to percutaneous pleural drainage. Esophagographic studies may confirm the presence of the EPF and demonstrate its site, although rarely indicated in this CT era. Ultrasound is helpful in detecting the hydro- or hydropneumothorax.

Management of the EPF depends on site, size, duration, and severity of perforation. Another important prognostic factor is extent of mediastinal involvement, which is better evaluated by chest CT. Conservative therapy includes drainage of the empyema, local irrigation, tube feeding, gastrostomy, or jejunostomy.<sup>[4]</sup> It is followed by definite surgery which includes repair or direct reconstruction of the esophagus. Early diagnosis and management of the EPF is important as it carries poor prognosis.

The index case presented with short history of retrosternal chest pain and vomiting. The differential causes of this nonspecific presentation include aspiration pneumonia, spontaneous pneumothorax, pulmonary emboli, myocardial infarction, aortic dissection, or intra-abdominal abnormalities such as cholecystitis, and gastritis. Chest CT showed EPF with collapse-consolidation

of right lower lobe and pleural thickening which may be attributed to infection of the airspace or pleural cavity. To conclude, spontaneous development of EPF is unusual entity with nonspecific clinical presentation. Chest CT is very useful modality for early diagnosis and management of the EPF. It should be done in patients with pleural effusion presenting with nonspecific clinical symptoms before any intervention or drainage.

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