

CASE REPORTS

SPONTANEOUS ESOPHAGOPLEURAL FISTULA IN A HIGH RISK INDIVIDUAL (TRUMPET BLOWER)

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Abstract:

A middle aged male patient, trumpet blower by occupation presented with progressive dyspnea with chest heaviness for 2 weeks and low grade fever for 1 week. It started with sudden severe retrosternal chest discomfort and episodic vomiting during practicing trumpet blowing. Patient was toxic with high fever, tachycardia and tachypnoea. Clinicoradiologically patient was diagnosed as left sided hydropneumothorax. Intercostal chest tube was inserted at left 5th intercostal space at midaxillary line following which pus and air came out. Pleural fluid was acidic with high amylase level and polymicrobial growth. Repeat chest x-ray showed partial resolution of left sided hydropneumothorax. From 4th day food particles were noticed in the drainage bag. For screening 0.1% sterile methylene blue was given orally which came in ICD tube and bag within 24 hours. Subsequently barium swallow esophagus, CT thorax with oral and intravenous contrast and upper gastrointestinal endoscopy confirmed left sided esophagopleural fistula.

Key words: Esophagopleural fistula, Trumpet blower, Esophageal perforation.

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Introduction:

Esophagopleural fistula (EPF) is an abnormal communication between two anatomically proximal structures so that contents of esophagus are drained into the pleural space with presentation mimicking primary respiratory disorder. EPF itself is an uncommon presentation, whereas spontaneous occurrence without any triggering factor is quite rare entity. Spontaneous esophageal rupture is difficult to diagnose and often mistaken for an respiratory and abdominal catastrophe. A high index of suspicion is necessary for diagnosis. Unless and until the primary disorder (esophageal perforation) is managed, significant morbidity and mortality is inevitable.

Case report:

A 52 years old smoker nondiabetic normotensive male patient, trumpet blower by occupation, residing at a rural part of West Bengal presented at our outpatient department with complaints of gradually progressive shortness of breath along with heaviness of chest for 2 weeks and low grade fever for last 1 week. Patient's illness started with a

sudden episode of severe retrosternal chest discomfort with radiation towards back along with episodic vomiting about 2 weeks back noticed during practice session of his trumpet blowing, which resolved to some extent within 2-3 days. Initial chest discomfort was followed by gradually progressive shortness of breath (modified medical research council mMRC grade 3 at presentation). There was no diurnal variation of dyspnoea, no history of wheeze, allergic rhinitis or suggestive features of paroxysmal nocturnal dyspnoea and orthopnoea. Patient also had intermittent low grade fever, often associated with episodes of chill and rigor and relieved on antipyretic medication.

General examination revealed raised temperature (102^oF), tachycardia (112/minute), tachypnoea (36/min), no clubbing, and no peripheral lymphadenopathy. On respiratory system examination, there was stony dull percussion note in left interscapular, infrascapular, infraaxillary and inframammary area with resonant note over remaining areas of left

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hemithorax. Breath sound was absent over the entire left hemithorax. The percussion was suggestive of a horizontal fluid level with presence of shifting dullness.

Routine blood examination revealed hemoglobin 9.3 gm/dl, total leucocyte count 16200 cells/mm³ with 88% of lymphocytes. Chest X-ray (CXR) showed homogenous opacity involving left hemithorax with air fluid level with contralateral mediastinal shift suggestive of left sided hydropneumothorax (figure 1). Aspirated pleural fluid was exudative with cell count >1000/mm³, all cells were lymphocytes, pH 6.8, amylase level 1840 IU/ml and culture showed growth of e.coli and pseudomonas. Intercostal chest tube (ICD) was inserted urgently at left 5th. intercostal space at midaxillary line following which turbid pleural fluid and air came out and patient improved symptomatically to some extent. But clinicoradiologically lung failed to expand and repeat CXR showed partial resolution of left sided hydropneumothorax (figure 2). Whereas from 4th. day onward we noticed drainage of food particles (mainly rice) in the drainage bag and tube. As the pH was acidic, amylase level was very high without

neutrophilia and food particles was present in pleural fluid, we tried to rule out any communication between pleura and esophagus.

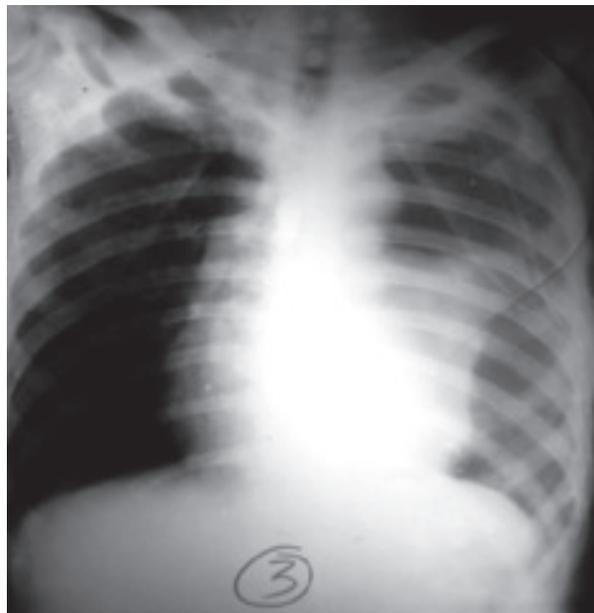


Fig. -2: Chest X-ray taken 3 days after intercostals tube insertion showing partial resolution of left sided huydropneumothorax with tube in situ.

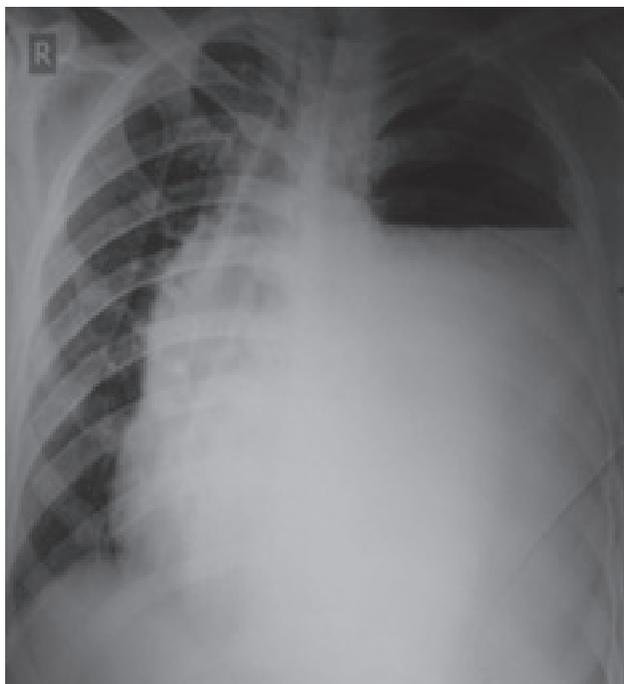


Fig.-1: Chest X-ray (PA view) showing homogenous opacity involving left hemithorax with air fluid level with contralateral mediastinal shift suggestive of left sided hydropneumothorax.

For screening we have performed methylene blue dye test where 0.1% sterile methylene blue was ingested orally by the patient which eventually came out in the ICD tube and bag within 24 hours. Barium swallow esophagus showed extravasation of radioopaque dye from lower part of esophagus into the left pleural cavity. Contrast enhanced CT thorax with both oral and intravenous contrast was done which also suggested a fistulous tract between esophagus and left pleural cavity supported by leakage of oral contrast in left sided pleural space (figure 3,4) with evidence of pleural effusion on the right side (which was later proved to be transudate). An upper gastrointestinal endoscopy revealed a small fistulous opening at lower part of esophagus with edematous surrounding mucosa.

Henceforth the final diagnosis was left sided spontaneous esophagopleural fistula. We had already given the intercostal tube which was repositioned later to facilitate the proper drainage from pleural space. Patient was put on ryles tube feeding to bypass the fistulous tract to promote healing of the fistula and reduction of surrounding mucosal edema so that we can proceed for definitive surgery. Thereafter

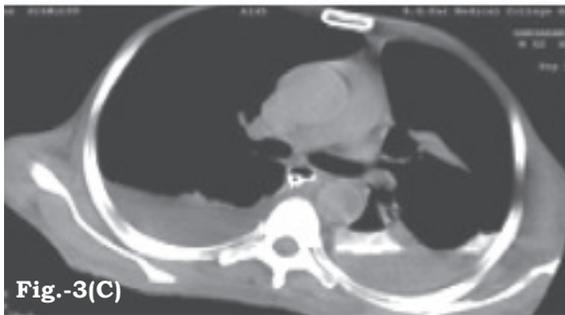
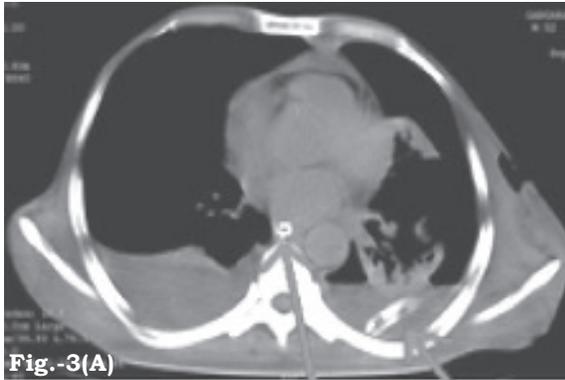


Fig.-3: CT scan thorax with oral and intravenous contrast showing (A) contrast within the esophageal lumen (B) initiation of leakage of oral contrast in the pleural space (C) partial leakage of oral contrast into the pleural space.

patient was referred to our surgical department where esophageal tear was repaired with omental patch and feeding jejunostomy was done (figure 5). Postoperatively patient improved with gradual expansion of lung. Patient was allowed oral feeding after 4 weeks and ICD tube was removed around 6 weeks later. Patient symptomatically improved with no evidence of reappearance of pleural effusion clinicoradiologically (figure 6). He was advised to quit his job of trumpet blowing as it could have been a triggering factor behind esophageal rupture. He is on regular follow up at our hospital and responding well.

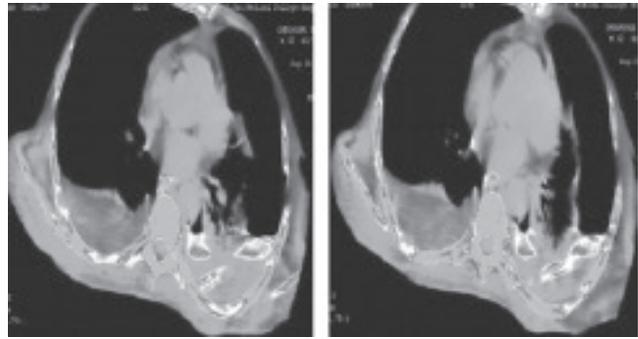


Fig.-4: Reconstituted CT thorax image showing oral contrast extravasation in pleural space.



Fig.-5: Postoperative image of the patient with intercostals tube inserted at left 5th intercostals space and drainage bag kept in situ following repair of esophageal tear.

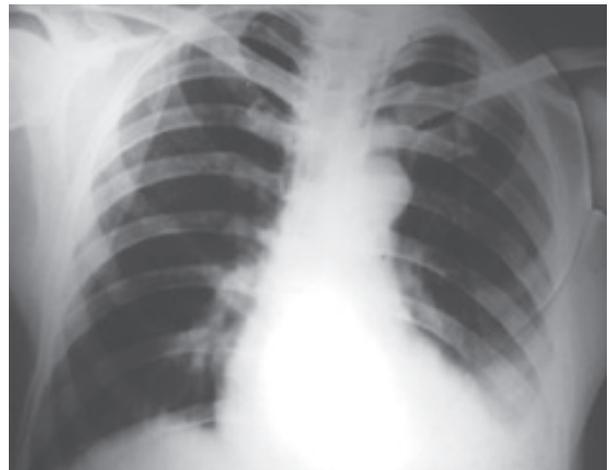


Fig.-6: Current chest X-ray (PA view) showing left sided pleural thickening with rib crowing with near total expansion of left lung.

Discussion:

Esophagopleural fistula (EPF) is one of the most common form of esophagorespiratory fistula that occur secondary to esophageal perforation.¹ The perforation most commonly arise due to esophagoscopy examination², remaining being foreign bodies (fishbone), carcinoma, gastric intubation, chest trauma and chest operations.³ But rarely spontaneous rupture of esophagus (Boerhaave's syndrome) may occur due to sudden rise of intraesophageal pressure (by contraction of cricopharyngeus muscle and closing of pyloric sphincter) associated with forceful vomiting or retching,⁴ classically after overeating or excessive drinking.⁵ A minor variety (Mallory Weiss tears) are mucosal tears caused by forceful or long term vomiting, retching or coughing and usually heal spontaneously. Characteristically this spontaneous rupture almost always involve the lower esophagus just above the diaphragm.³

Esophageal rupture occur due to a momentary rise of intragastric pressure (as in vomiting, trauma, seizure or other straining) that overcome the tensile strength of normal esophageal wall whereas accompanying relaxation of the lower esophageal sphincter permit gastric contents to enter and distend the lower esophagus. If the upper sphincter does not open to allow regurgitation of esophageal content, then rising intraesophageal hydrostatic pressure may rise to such a point that its wall may give away at its weakest point, usually in the lower left posterolateral wall where the muscle bundles in the longitudinal layer of esophagus separate, allowing a bubble of mucosa to protrude into the mediastinum and burst.⁵ That may also explain the occurrence of resultant pleural effusion on the left side, though it may be right sided or bilateral.

Spontaneous esophageal rupture is difficult to diagnose and often mistaken for an abdominal catastrophe.⁵ A high index of suspicion is necessary to diagnose spontaneous esophageal perforation with resultant complications:-

A) excruciating retrosternal chest pain with a sensation of tearing or bursting in the lower part of chest or the epigastrium, often unrelieved by opiates⁶
 B) may have preceding history of vomiting, dysphagia or associated with hemoptysis, dyspnea
 C) clinically subcutaneous emphysema that appear first in the suprasternal notch suggest esophageal perforation^[6]
 D) chest radiograph revealing pleural effusion (60%) frequently hydropneumothorax (25% cases)^[7] particularly when the mediastinal pleura rupture, diffuse mediastinal widening with air visible within

the mediastinal compartments and soft tissues (hallmark sign)^{3,5} E) chest CT scan showing paraesophageal air tracks⁸ F) characteristic pleural fluid picture revealing high amylase level (best indicator of esophageal rupture), mainly salivary variant rather than pancreatic variety⁹, low pH (pH < 7.00 suggest increased likelihood of rupture, whereas pH < 6.00 is highly suggestive of esophageal rupture^[10]), presence of squamous epithelial cells (by Wright's stain)^[11], presence of ingested food particles (virtually diagnostic) and multiple pathogens on smear or culture (polymicrobial empyema particularly when the daily pleural fluid output is high)³

However for confirmation, following imaging modalities may be contemplated-

A) contrast study of the esophagus (with barium or gastrograffin or meglumine or ioxaglate sodium) in decubitus position which is positive in approximately 85% of patients^[12] demonstrating the actual site of perforation and its interconnecting cavities
 B) chest CT with oral and intravenous contrast showing extravasation of orally ingested contrast material into the periesophageal or pleural space (pathognomonic sign)^[13] extraluminal air and focal thickening or ballooning and thinning of the esophageal wall at the site of perforation
 C) upper gastrointestinal (g.i.) endoscopy
 D) passage of orally ingested colored dye/contrast agent (sterile methylene blue used in our case) in the intercostals drainage bag may be an effective intervention to stamp the diagnosis.

We had checked and reviewed the role and safety of methylene blue in gastrointestinal tract procedures. Sterile solution of methylene blue can be given via nasogastric tube into the gastric pouch to test the integrity of anastomosis in nissen fundoplication.¹⁴ It may also be instilled into the esophagus in an attempt to localize the injury of cervical esophagus.^[15] Its use is also well documented in upper g.i. endoscopy in various conditions like Barrett's esophagus, early gastric carcinoma.

The treatment of choice is exploration of the mediastinum with primary repair of esophageal tear and drainage of the pleural space and mediastinum. Associated conservative treatment consisting of antibiotic and nasogastric suction may be performed. However EPF without mediastinal involvement usually carries good prognosis and may respond to percutaneous pleural drainage.

Our case was interesting in the perspective of following factors-

a) bilateral involvement (though fistulous tract with leakage of oral contrast was seen only on left

pleural space, whereas right sided effusion was transudative)

- b) spontaneous occurrence without any definite triggering factors
- c) occurrence in a high risk individual (trumpet blowing warrant repeated episodes of sudden transient rise of intraesophageal pressure which may predispose to esophageal rupture)
- d) use of sterile methylene blue as a simple cost effective screening tool for diagnosis of fistula
- e) early intervention and coordinated management strategy ensuring patient survival with minimal morbidity.

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