



Case report

Tension hydropneumothorax as the initial presentation of Boerhaave syndrome

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ABSTRACT

Boerhaave syndrome, a rare yet frequently fatal diagnosis, is characterized by the spontaneous transmural rupture of the esophagus. The classic presentation of Boerhaave syndrome is characterized by Mackler's triad, consisting of chest pain, vomiting, and subcutaneous emphysema. However, Boerhaave syndrome rarely presents with all the features of Mackler's triad; instead, the common presentation of Boerhaave syndrome includes chest or epigastric pain, severe retching and vomiting, dyspnea, and shock. These symptoms are typically misdiagnosed as cardiogenic in origin. Due to its atypical presentation, rarity, and mimicry of emergent conditions, diagnosis of Boerhaave syndrome is often delayed, resulting in a high mortality rate at the time of diagnosis and with a subsequent exponential increase in mortality if treatment is delayed by greater than 48 hours. Here, we report two atypical presentations of Boerhaave syndrome presenting as tension hydropneumothorax and review ten previously reported cases of Boerhaave syndrome presenting as tension hydropneumothorax. This review serves to raise clinician awareness about the expansive and elusive ways by which esophageal perforation may present, and thereby facilitate timely and potentially life-saving diagnosis.

1. Introduction

Spontaneous transmural rupture of the esophagus, or Boerhaave syndrome, was first described in 1724 by Hermann Boerhaave [1]. Mackler's triad, consisting of chest pain, vomiting, and subcutaneous emphysema represents a classic presentation. However, Mackler's triad is only present in about 5% of case, with chest pain being the most common feature [2,3]. Boerhaave syndrome itself is rare, with an annual incidence of 3.1 per 1,000,000 [4]. Coupled with its mimicry of other conditions, the diagnosis of Boerhaave syndrome is often delayed, with mortality rates up to 30% at the time of diagnosis that significantly increase by treatment delays > 48 hours [5]. We report two cases of Boerhaave syndrome with tension hydropneumothorax as the initial presentation and review other reported cases.

2. Case presentations

2.1. Case 1

A 65-year-old male with hepatitis C, alcoholism and prior small

bowel obstruction (SBO) from abdominal surgery was admitted with 3 days of abdominal pain, emesis and recurrent partial SBO. The admission chest x-ray (CXR) was unremarkable. On hospital day 2, he developed acute chest pain with electrocardiogram demonstrating ST-segment elevation. Emergent cardiac catheterization revealed no obstructive coronary artery disease. Post procedure, he developed excruciating chest pain radiating to his back with tachycardia, hypotension and absent breath sounds over the left lung. CXR demonstrated interval opacification of the left hemithorax with mediastinal shift. An emergent needle thoracostomy produced air and dark fluid containing food debris, followed by tube thoracostomy which drained 4 L of bilious, enteric output. Chest computerized tomography (CT) demonstrated a large left hydropneumothorax with free air around the esophagus (Fig. 1). Esophagogastroduodenoscopy demonstrated “black esophagus” in a circumferential distal third of the esophagus consistent with necrosis and perforation. He underwent emergent surgery with cervical esophagostomy, laparotomy, and gastrostomy tube placement. His post-operative course was complicated by hypoxemic respiratory failure and on hospital day 21, massive gastrointestinal hemorrhage and death.

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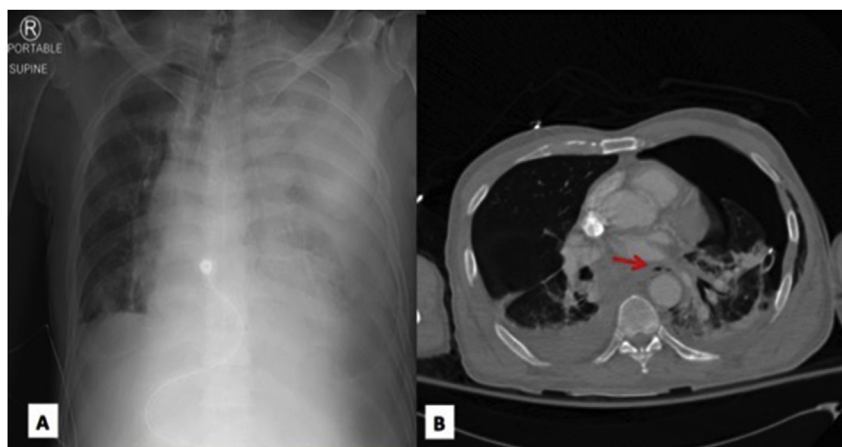


Fig. 1. A) AP Chest radiograph demonstrating complete opacification of the left lung with mediastinal shift to the right and a small right-sided pleural effusion. B) Chest CT demonstrating extra-luminal free air in the mediastinum (red arrow), left-sided hydropneumothorax and small right-sided pleural effusion. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

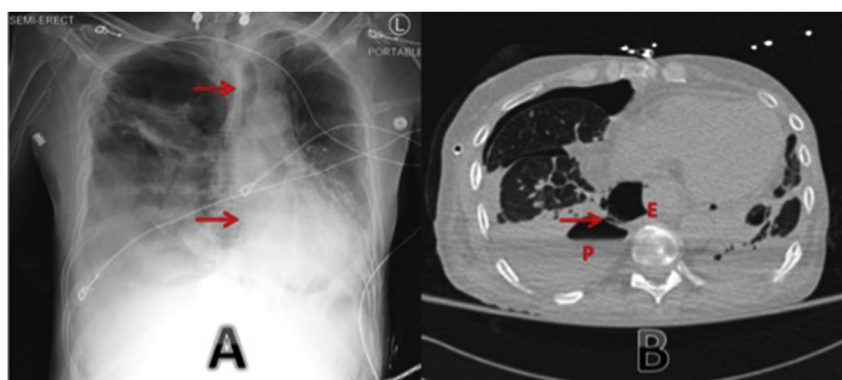


Fig. 2. A) AP Chest radiograph demonstrating a right-sided tension hydropneumothorax with mediastinal shift to the left (red arrows). B) Chest CT demonstrating free air around the esophagus (E) communicating (red arrow) with the pleural space. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

Table 1
Presenting symptoms of tension hydropneumothorax in Boerhaave syndrome.

Symptoms	% of patients presenting with symptoms
Chest pain	6/12 (50%)
Epigastric pain	4/12 (33%)
Vomiting	11/12 (92%)
Dyspnea/Tachypnea	8/12 (67%)

2.2. Case 2

A 60-year-old male with alcoholism and chronic dysphagia secondary to Barrett's esophagus and achalasia was admitted with alcohol withdrawal and aspiration pneumonia. Notably, he had severe dysphagia for which he would self-induce vomiting to alleviate sensations of choking and alleviate hiccups. He failed a swallow evaluation and nasogastric tube placement was attempted but was unsuccessful. On hospital day 10, he developed tachycardia, tachypnea, hypoxemia, and chest pain, with opacification of the right hemithorax with mediastinal shift (Fig. 2). An emergent needle decompression was performed, followed by tube thoracostomy with drainage of over 1 L of exudative fluid (pH of 6.9, amylase > 6000 U/L). Chest CT confirmed free air around the esophagus in communication with the pleural space consistent with esophageal perforation. He underwent surgical repair with clinical improvement. Patient remained hospitalized undergoing physical therapy, but developed a fatal intracranial bleed on post-operative day 30.

3. Discussion

The classic presentation of Boerhaave syndrome includes chest or epigastric pain (83%), severe retching and vomiting (79%), dyspnea

(39%), and shock (32%) [6]. Esophageal rupture occurs when an abrupt increase in intraluminal esophageal pressure forces gastric contents against the cricopharyngeal muscles causing transmural esophageal rupture [7]. As the esophagus ruptures, the parietal pleura can also rupture or become breached by gastric enzymes, leading to pneumothorax or hydropneumothorax. If the esophageal perforation is sealed, patients may appear deceptively well without symptoms and/or hemodynamic instability [6].

Boerhaave syndrome presenting as tension hydropneumothorax is an extremely rare phenomenon. A MEDLINE/PubMed and Google Scholar search with search terms of “Boerhaave syndrome”, “esophageal rupture”, “esophageal perforation”, “tension hydropneumothorax”, “tension pneumothorax”, and “pneumothorax”, only identified 10 reports of tension hydropneumothorax in Boerhaave syndrome in addition to our two cases.

In patients with tension hydropneumothorax as the initial presentation of Boerhaave's syndrome, the most common symptoms were recent vomiting (11 of 12), dyspnea (8 of 12), chest pain (6 of 12), and epigastric pain (4 of 12) (Table 1). Mackler's triad of chest pain, vomiting, and subcutaneous emphysema was not seen in any of the 12 cases in our series, suggesting the classical teaching of Mackler's triad is neither sensitive nor specific for this diagnosis.

The non-specific presenting symptoms and mimicry of other emergent conditions often require additional studies to establish the diagnosis. In these 12 cases, the initial imaging modality was a CXR, which revealed tension hydropneumothorax – a rare presenting sign for esophageal rupture (Table 2). Boerhaave syndrome may also be suspected based on mediastinal widening, pneumothorax, hydrothorax, hydropneumothorax, mediastinal shift, and/or subcutaneous emphysema [8]. Esophagram or CT scan with water-soluble oral contrast is the gold standard for diagnosing esophageal perforation [2,3]. However, due to the high false negative rate of the contrast esophagram (15–25%) and

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