

# Bee Sting to Boerhaave's Syndrome

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## ABSTRACT

Boerhaave's syndrome is a rare condition defined as the spontaneous rupture of the esophagus that generally occurs due to retching, forceful vomiting and sometimes even spontaneously. Atypical presentation often misleads the diagnosis leading to a delay in early intervention, and a strong clinical suspicion is indeed required to diagnose the condition. Definitive treatment being surgical repair, endoscopic intervention can be attempted in nonseptic patients.

**Keywords:** Acute hypoxemic respiratory failure (AHRF), Anaphylactic reaction, Esophageal injury, Pneumothorax.

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## INTRODUCTION

We present a case of 39-year-old male patient admitted following bee sting injury, who was treated initially as anaphylactic shock and myocardial infarction. As the patient had retching, vomiting initially following bee sting injury and as he did not improve with the conventional management strategies of anaphylactic shock and myocardial infarction, clinically suspected as esophageal rupture, because he developed pneumothorax on day 2 of hospitalization and persistent drain output following tube thoracostomy.

Initial upper gastrointestinal endoscopy did not reveal perforation of the esophagus. A strong clinical suspicion, elevated pleural fluid amylase levels, and leakage of oral contrast into the pleural cavity in CT of chest and abdomen established the diagnosis of Boerhaave's syndrome. Although surgical intervention is definitive, in our case, the patient improved following endoscopic intervention and he was discharged home in stable condition.

## CASE DESCRIPTION

A 39-year-old male patient presented with alleged history of bee sting injury over the face and trunk following which he developed edema of the face and neck. He had a history of retching, vomiting a few hours following aforementioned event, and he developed dyspnea with chest discomfort. He was evaluated elsewhere, where he received steroids and bronchodilators for suspected anaphylaxis. In view of worsening symptoms, he was referred to our hospital for further management.

On arrival to the emergency department, he was tachypneic, hypoxemic, and tachycardic and was in severe respiratory distress with oxygen saturation of 80% on room air. His trachea was emergently intubated in view of worsening hypoxemia despite oxygen support and severe respiratory distress and vasopressor support initiated to maintain the hemodynamics. After initial stabilization in the emergency department, he was shifted to critical care unit for continuation of care.

Physical examination revealed edema of the face and neck regions with bilateral crackles. In view of chest pain and lung signs, cardiologist opinion was sought, but ECG and echocardiography were normal. Arterial blood gas analysis revealed metabolic and respiratory acidosis with dyselectrolytemia, and serum urea and creatinine levels were elevated, which were

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managed conservatively. Laboratory evaluation was suggestive of hemoconcentration with the rest of the parameters within normal limits. Hypoxemia persisted despite ventilatory support, and clinical examination revealed decreased air entry on the left side and increasing subcutaneous emphysema over the neck. Diagnosis of pneumothorax was made, which was supported by absent lung sliding and lung point on ultrasonography of chest and pneumothorax on roentgenogram of chest on the left side. Tube thoracostomy was done for left pneumothorax and drained 650 mL of blackish brown fluid, which was sent for analysis.

HRCT chest was done suggestive of pneumomediastinum with right pleural effusion for which right tube thoracostomy was done, which drained 350 mL of brownish black fluid. Pleural fluid analysis was suggestive of exudative collection with total count of 8000 cells with neutrophil predominance of 80% and increased lactate dehydrogenase levels. Medical gastroenterology consultation was taken. The upper gastrointestinal endoscopy was performed but inconclusive.

By the third day of hospitalization, the roentgenogram of chest improved, the patient was normoxemic, and hemodynamics was stabilized. He was weaned off the vasopressor support and the ventilator support and eventually got extubated. In view of persistent thoracostomy drain on the left side, contrast-enhanced CT of chest and abdomen with oral contrast was performed. During

procedure, there was a gradual accumulation of contrast into the left pleural cavity and draining into the left thoracostomy drain. Pleural fluid amylase was grossly elevated to 55,324 units/dL. Hence he was provisionally diagnosed as having esophageal rupture and planned for surgical repair or endoscopic stent placement.

Repeat upper gastrointestinal endoscopy revealed full-thickness perforation at the hiatal pouch on the gastric side with normal stomach and duodenum. The perforation was feeding into the pleural cavity and filled with purulent secretions. An over-the-endoscopy clip was placed immediately to seal the perforation, and self-expandable metallic stent placement was planned later as a staged procedure. Postprocedure immediate contrast study did not reveal any leak with free flowing of contrast into the stomach.

The patient general condition gradually improved with decreasing drain in the right thoracostomy tube, and hence, the drain was removed. Gastrograffin study was scheduled after 48 hours to detect any postprocedure leak. The patient was eventually discharged in hemodynamically stable condition to another hospital. The patient was being followed up later with us and there was no gastrograffin leak into the pleural cavity on check upper gastrointestinal endoscopy with intact sealed perforation. He was started on oral liquids and discharged home in stable condition.

## DISCUSSION

Boerhaave's syndrome is defined as the spontaneous rupture of the esophagus following a sudden increase in intraesophageal pressure combined with negative intrathoracic pressure such as that associated with severe straining or vomiting, and less frequently with childbirth, seizure, prolonged coughing or laughing, or weight lifting, which results in a longitudinal esophageal perforation. It most often occurs in the distal posterolateral aspect of the esophagus.<sup>1</sup> Boerhaave's syndrome is more common in males.

Esophageal perforations are rare, with an incidence of 3.1 per 1,000,000 per year.<sup>2</sup> Among esophageal perforations, approximately 15% are spontaneous perforations.<sup>3</sup> Meckler's triad of vomiting, pain, and subcutaneous emphysema are the characteristic features of Boerhaave's syndrome.<sup>4</sup>

Many a time, they present with typical signs and symptoms often misleading the diagnosis and delaying the appropriate intervention. They are managed initially as acid peptic disease, pancreatitis, myocardial infarction, tension pneumothorax, or an acute aortic dissection.

In our case, the patient initially presented with anaphylaxis and was being evaluated and treated as anaphylactic shock and myocardial infarction. As the patient remained hypoxemic with gradually increasing subcutaneous emphysema, pneumothorax was contemplated, which was supported by ultrasonography of chest and roentgenogram of chest, and tube thoracostomy was done on the left side. The pleural fluid initially was brownish black in appearance, and since occurred in the context of severe retching, vomiting following bee sting injury, an esophageal rupture was suspected, and hence, pleural fluid analysis was sent.

Initially, the pleural fluid analysis was exudative, but drain output persisted without any signs of fever or increasing total leukocyte count and stable hemodynamics. This raised the suspicion of esophageal perforation without mediastinitis, and hence, pleural fluid amylase analysis was done, and it was grossly elevated. Initial upper gastrointestinal endoscopy was inconclusive. O'Kelly et al. published a case report on Boerhaave's syndrome where perforation only became evident following air insufflation at

endoscopy.<sup>5</sup> To establish the diagnosis, CT of chest and abdomen with oral contrast was performed in which the contrast was feeding from the esophagus into the pleural cavity, and this confirmed the diagnosis of Boerhaave's syndrome. Immediately repeat upper gastrointestinal endoscopy was performed, which revealed full-thickness perforation in the hiatal pouch and sealed endoscopically.

Boerhaave's syndrome has a high mortality because of mediastinitis, sepsis, and septic shock. Roentgenogram of chest with contrast-enhanced CT of chest and abdomen is of great value in diagnosing Boerhaave's syndrome. Pleural fluid amylase analysis can be used as an initial investigation to narrow down the differential diagnosis and assist in the diagnosis of Boerhaave's syndrome.

Management of esophageal perforations can be primarily conservative, endoscopic, or surgical.<sup>6</sup> Rapid surgical intervention is widely recommended, with primary repair where possible.<sup>7</sup> Esophageal replacement is done after 6 weeks, usually with the colon or the stomach.<sup>8</sup> On the contrary, other authors<sup>9</sup> advocated that reinforced primary repair can be done for most late perforations. Jougon et al. even advocated that all esophageal perforations can have primary repair no matter the time between perforation and treatment.<sup>10</sup> Endoscopic repair of Boerhaave's perforations with a self-expanding metallic stent placement can be done in patients without SIRS and septic shock. Dickinson et al.<sup>11</sup> published a 10-year review on endoscopic repair of Boerhaave's perforations and concluded that it can be useful in carefully selected patients without evidence of systemic sepsis.

## CONCLUSION

Our case highlights the varied presentation of this potentially fatal condition. This patient initially presented with signs of anaphylaxis, after honey bee sting. We had a high index of suspicion, as this patient had bilateral pleural effusion with increased drain amylase and exudative pleural effusion. The quick diagnosis is decisive for the outcome of the treatment of patients with esophageal perforation because of the serious complications such as mediastinitis, empyema, and sepsis. Endoscopic technique can provide a less morbid therapeutic intervention as in this patient the leak is contained between the mediastinum and visceral lung pleura.

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