

Boerhaave's Syndrome Presenting as a Right-sided Pleural Effusion

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Abstract

Introduction: Boerhaave's syndrome is an uncommon condition where there is oesophageal rupture following forceful vomiting, subsequent mediastinitis and is associated with high mortality and morbidity in the absence of therapy. We present a case of Boerhaave's syndrome in a 79-year-old woman who developed a right-sided effusion, an unusual presentation. **Clinical Picture:** A 79-year-old woman developed a right-sided empyema and mediastinitis after a bout of repeated vomiting. Gastrograffin swallows and oesophago-gastro-duodenoscopy confirmed oesophageal rupture. **Treatment:** The patient was treated with antibiotics and a tube thoracostomy. An open thoracotomy, oesophagostomy and thoracic window was subsequently performed. **Outcome:** The oesophageal rupture was contained but patient died from postoperative complications. **Conclusions:** Physicians should have a high index of suspicion of oesophageal rupture when patients present with Meckler's triad of symptoms and especially when pleural fluid cultures grow bacteria native to the gastrointestinal tract.

Ann Acad Med Singapore 2003; 32:415-7

Key words: Boerhaave's syndrome, Empyema, Mediastinitis, Oesophageal perforation

Case Report

A 79-year-old Chinese woman with no significant past medical history was admitted to another hospital with symptoms of abdominal pain and haemetemesis of approximately 200 mL of blood after a severe bout of vomiting. She declined a oesophago-gastro-duodenoscopy (OGD) and was clinically diagnosed to have Mallory-Weiss syndrome. A chest radiograph on admission showed a right pleural effusion. Pleural aspiration under ultrasonic guidance revealed blood-stained pleural fluid. Results of her pleural fluid studies are as follows: red blood cells (RBC), 5 per high power field (hpf); white blood cells (WBC), 8 per hpf; lactate dehydrogenase (LDH), 311 U/L; and glucose 0.9 mmol/L. She was diagnosed to have a right parapneumonic effusion secondary to aspiration pneumonia and was treated with oral antibiotics. As her abdominal pain and haemetemesis resolved, she was discharged at her request. The pleural fluid cultures subsequently grew *Candida* and *Lactobacillus*.

She presented to our institution 7 days later with vague abdominal pain not associated with vomiting, constipation or diarrhoea. Clinically, she was afebrile and had negative abdominal findings on palpation. On auscultation of the

lungs, she had decreased air entry and crepitations in the right hemithorax posteriorly. A chest radiograph showed a right pleural effusion (Fig. 1). Computed tomographic (CT) scan of the thorax showed a small loculated right effusion and enhancing visceral, and parietal pleura suggestive of an empyema. There was also free air in the mediastinum with associated mediastinitis (Fig. 2). A small-bore pleural catheter was inserted under radiological guidance. The fluid drained was exudative with high white counts and extremely low glucose levels. Results of the pleural fluid studies are as follows: pH, 5.7; total protein, 66 g/dL versus serum total protein 62 g/dL; glucose, 0.6 mmol/L; and LDH, 33,905 U/L. Culture of the fluid grew *Candida tropicalis*, *Candida glabrata* and *Klebsiella*. She was treated with intravenous fluconazole and piperacillin/tazobactam. As she was suspected to have an oesophageal rupture, a gastrograffin swallow was performed which showed a dilated retrocardiac portion of the oesophagus with leakage into the mediastinum and the right pleural cavity (Fig. 3). An OGD done the following day showed a large oesophago-pleural fistula measuring 5 cm in diameter at the 26 cm mark. No tumour was seen.

Thoracotomy and oesophagectomy was attempted but as

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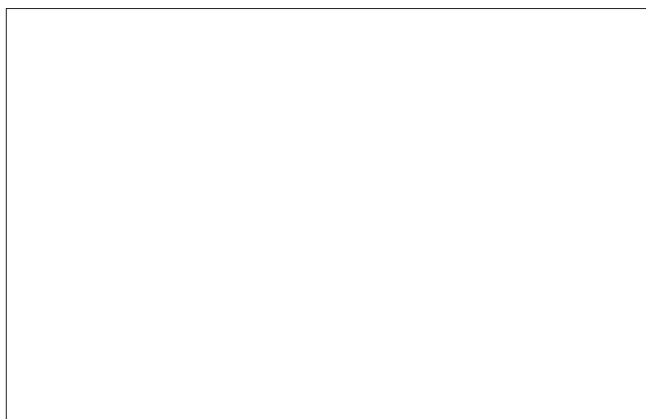


Fig. 1. Chest radiograph showing a right pleural effusion.

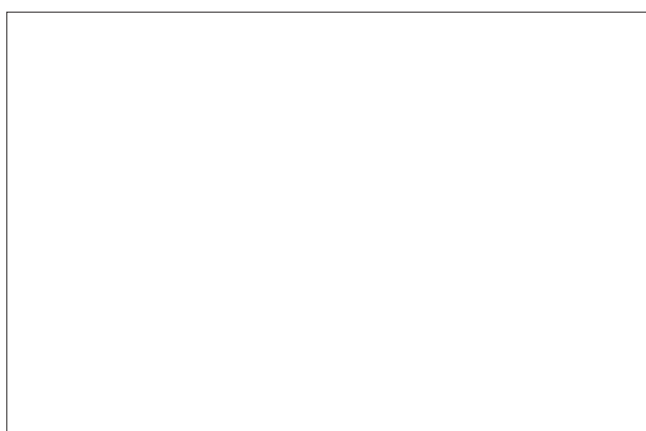


Fig. 2. CT scan of the thorax showing a loculated right effusion and enhancing visceral and parietal pleura. Free air is seen in the mediastinum.

she was unable to tolerate single-lung ventilation, the affected pleural space was irrigated to remove the copious amounts of pus and a thoracic window was created. A feeding jejunostomy and cervical oesophagostomy was fashioned. Her postoperative period was stormy, complicated by septicaemic shock from nosocomial pneumonia and Methicillin-resistant *Staphylococcus aureus* (MRSA) wound infection, disseminated intravascular coagulation and acute renal failure. She subsequently succumbed to multi-organ failure 27 days following surgery.

Discussion

Atrocis, nec descripti pruis, morbid historica was coined by Dr Herman Boerhaave (1669-1738), a professor of medicine, botany and chemistry, in Leiden, Holland in 1724. He had performed an autopsy on Baron John van Wassenaar of the Dutch Navy after the latter died suddenly following a severe bout of retching and found a large perforation on the left side of the oesophagus about 3 times the size of his thumb.

Since then, Boerhaave's syndrome has been used to describe full thickness perforations of the oesophagus not

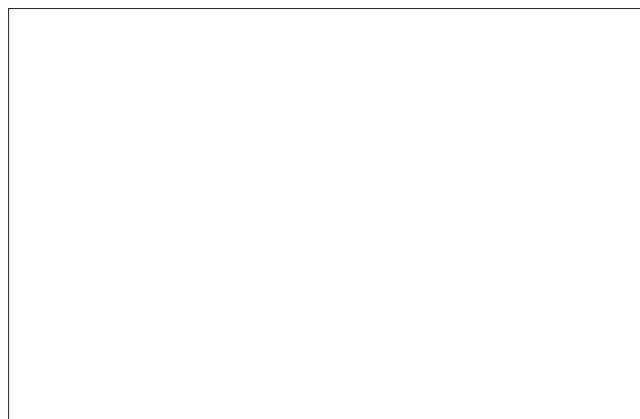


Fig. 3. Gastrograffin swallow showing a dilated retrocardiac portion of the oesophagus with leakage into the mediastinum and the right pleural cavity.

associated with external trauma, instrumentation or foreign bodies. Incidence rates have been described to occur between 1 in 8000 and 1 in 20,000 hospital admissions with a mortality rate of 18% to 29%. Mortality and morbidity increase in direct proportion to the time between diagnosis and appropriate surgical intervention.

The mechanism of rupture as elucidated by Rogers and associates¹ is an increase in intragastric pressure coupled with opening of the lower oesophageal sphincter to allow entry of gastric contents into the lower oesophagus. If the upper sphincter does not open, hydrostatic pressure within the oesophagus rises to a point when the wall gives way at its weakest point: the posterolateral wall on the left. This is where muscle bundles in the longitudinal layer of the oesophagus separate, and leads to protrusion of mucosa into the mediastinum and subsequent rupture. Lacerations are usually vertically orientated and within 4 cm of the gastro-oesophageal junction. Ninety per cent mortality rates have been reported if there is delayed diagnosis and treatment. In our patient, the rupture was right-sided which is a rare occurrence. Spillage of gastric acid and digestive enzymes into the mediastinum is potentially fatal in 100% of cases.

Clinically, patients with Boerhaave's syndrome, as described by Meckler,² are said to present classically with the triad of vomiting, lower thoracic pain and subcutaneous emphysema. Our patient had presented with vomiting and abdominal pain to 2 different institutions. However, Flynn et al³ reported that only one third of patients with Boerhaave's syndrome actually presented with Meckler's triad. Similarly, Lemke and Jagminas⁴ described that only a minority of patients admitted to hospital with a definite diagnosis of oesophageal rupture actually presented with the classic triad. In 1987, Graeber et al⁵ also found that patients who presented with Boerhaave's syndrome had a myriad of presentations outside Meckler's triad, such as fever and dysphagia.

Radiologically, Boerhaave's syndrome can present early with a widened mediastinum and pneumomediastinum on chest X-rays and CT of the thorax. Peri-oesophageal fluid, esophageal thickening and extraluminal contrast leak can also be seen. Eventually, pleural effusions and hydro-pneumothoraces develop. Barium swallow is the most common diagnostic test used to diagnose Boerhaave's syndrome, although barium is known to cause mediastinal fibrosis. Gastrograffin swallows have been suggested to be a safer and more reliable test to detect leaks from the oesophagus; but gastrograffin disperses quickly in the mediastinum and hence leaks may be missed.

Pleural fluid examination can be an adjunct to diagnosing Boerhaave's syndrome as described by Drury et al.⁶ Their patient had a negative barium swallow but subsequent pleural fluid testing showed an acidic pH with increased amylase of salivary origin. Pleural fluid cytology showed undigested food particles. It is suggested that pleural fluid be sent for cytology as smears would miss such important findings. Our patient had an empyema which was polymicrobial on culture on two occasions. In addition, organisms from the gastrointestinal tract were cultured at the first presentation. This should have raised the suspicion of an oesophageal rupture.

The role of OGD in the presence of oesophageal rupture is controversial. It has been suggested that endoscopy and insufflation of air can extend the perforation and also introduce air into the mediastinum. In this instance, an OGD would have been indicated, in view of the significant history of vomiting and haemetemesis. This would have led to an early diagnosis of the patient's problem and active management could have been instituted earlier.

Various management options are available, both surgical and non-surgical. However, there are no randomised controlled trials comparing the outcomes of surgical and non-surgical management of patients with Boerhaave's syndrome. Non-surgical management is suggested for patients who are diagnosed late, clinically stable and demonstrate tolerance to the perforation which is well contained. Such patients should be treated by nil by mouth with intravenous fluids, nasogastric drainage, tube thoracotomy, antibiotics and parenteral nutrition. There have been anecdotal reports of patients treated non-surgically with successful outcomes. One patient was described to be treated with parenteral nutrition for 25 days and discharged well after a 6-month hospital stay.⁷ Another patient was treated with parenteral nutrition for 4 months and discharged well from the hospital 7 months after admission.⁸

Surgical intervention which includes open thoracotomy, debridement and surgical closure of the perforation is recommended if the diagnosis is made early. Diversion surgery by oesophagostomy, oesophagectomy or T-tube diversion⁹ is usually reserved for critically ill patients who have extensive involvement of the oesophagus and mediastinum. Use of self-expanding metallic stents have been reported to be successful if the diagnosis is made within 24 hours.¹⁰

Conclusion

The clinical diagnosis of Boerhaave's syndrome is difficult as the presenting symptoms are myriad. Physicians should have a high index of suspicion when patients present with Meckler's triad. In addition, polymicrobial cultures from the pleural effusion, especially bacteria of gastrointestinal origin, should prompt the physician to the likely diagnosis of oesophageal rupture.

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