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Atypical chest pain: A case report of Boerhaave syndrome

Christelle Margot, Jérémy Desmercieres

ABSTRACT

The spontaneous rupture of the esophagus, commonly called Boerhaave syndrome, is a rare and serious disease with poor prognosis due to its diagnostic difficulty. The patients suffering from this condition need multidisciplinary technical platform, and the survival of patients correlates highly with early medical care with fluid resuscitation, antibiotics administration, and surgical care to treat the perforation using endoscopy or extrathoracic surgery. This article presents a case report of Boerhaave syndrome occurring in a Community Hospital and discusses the need for initial management and referral to University Hospital.

Keywords: Boerhaave, Esophagus, Rupture

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INTRODUCTION

Boerhaave syndrome is a rare gastrointestinal disease with high morbidity and mortality rate [1], described for the first time in 1724 [2]. It is due to spontaneous barotraumatic rupture due to sudden intra-esophageal hyperpressure during vomiting [3].

A meta-analysis of 2013 regroups 75 studies resulting with 2971 patients, and the authors conclude that studies may be biased by patient selection and limited experience due to the scarcity of this disease [4]. The lack of clinical specificity of this pathology can lead to a diagnostic and therapeutic delay which could be fatal for the patient [5]. Even though there is no standardized care, studies agree that the first 24 hours (golden 24-hours) are crucial for the prognosis [6]. Recent meta-analysis of 2020 regrouping 384 patients among 25 studies confirms that an early diagnosis (<24 hours) correlates with a better clinical outcome [7]. The management is more complicated in community hospitals where the diagnosis and therapeutic tools, as well as the specialists are not easily available. However, the initial stabilization with fluids and antibiotics is also essential for the outcomes of the patient, while waiting for the definitive medical or surgical treatment [8].

CASE REPORT

A 61-year-old male patient with a history of ascending aorta endoprosthesis and aortic valve replacement with a mechanical prosthesis anticoagulated with acenocoumarol was evaluated in the emergency department of a community hospital for chest pain. The pain started few hours ago, followed by several episodes of emesis. Despite his symptomatology he went to a fitness center and practiced weight lifting. After the effort the pain became more intense associated with episode of fainting.

On physical examination, blood pressure was 97/52, pulse rate was 133/min, respiration rate was 34/min, temperature was 37.1°C, and oxygen saturation was 91% on 15L of oxygen. Pulmonary examination revealed tachypnea with accessory muscles use, presence of subcutaneous emphysema, and absence of breath sound on the left chest. Abdominal palpation showed epigastric guarding. The remainder of the physical examination was unremarkable.

Laboratory studies revealed normal electrolytes, a serum creatinine of 107 umol/L, total leukocyte count of 18.5 × 10^9/L with neutrophilic predominance (86.4%), serum hemoglobin of 15.6 g/dL, platelet count within normal range, and international normalized ratio (INR) of 2.5. Arterial blood gas studies were initially normal without lactic acidosis.

A computed tomography scan (CT scan) showed esophagus rupture with pneumomediastinum associated with significant left pleural effusion, ipsilateral pneumothorax, and major stomach distention (Figure 1).

Evolution

Intravenous fluid resuscitation was initiated and a nasogastric tube was placed. A chest tube was inserted on the left side, with previous hemostasis reversion by administration of vitamin K, tranexamic acid, and prothrombin complex concentrate (PCC), allowing the evacuation of 1 L of blood with a positive effect on breathing. Prophylactic antibiotics composed by cefepime and vancomycin were initiated. Due to a decrease in hemoglobin values during a second blood test the patient received a red blood cells transfusion.

The patient was then transferred to the University Hospital of Lausanne in Switzerland where an urgent esophagogastroduodenoscopy (EGD) was performed and revealed a deep injury from the esophagogastric junction to around 43 cm, with muscularis tear, and small openings corresponding to the esophagus perforation. Two ulcers Forest III in the cardia with prolongation to the distal esophagus was also highlighted (Figure 2).

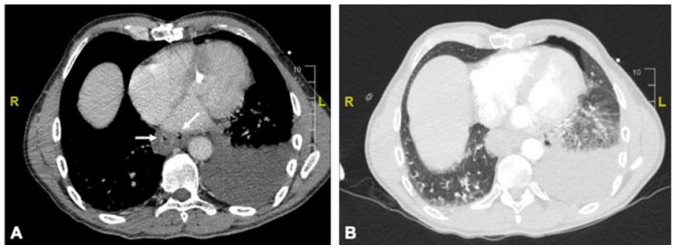

Figure 1: (A) Chest computed tomography scan with injection of iodinated contract: distal esophagus rupture with pneumomediastinum facing the left ventricle. (B) Chest computed tomography scan in pulmonary window: large left pleural effusion with small ipsilateral pneumothorax.

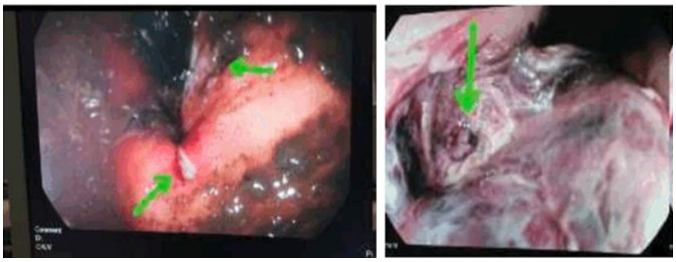

Figure 2: Esogastroduodenoscopy: Esophageal perforation with muscularis tear and presence of opening in the esophagogastric junction. Ulcer Forest III from the cardia to distal esophagus.

During this EGD a double esophageal prosthesis was placed with a nasojejunal tube.

The antibiotics were changed to piperacillin-tazobactam and antifungal therapy with fluconazole was also initiated. The prosthesis was removed at day 7 with an esosponge insertion at the level of the perforation. The esosponge was changed several times and definitely removed at day 16 due to good evolution.

The patient developed a persistent feverish state despite large spectrum antibiotics, with purulent discharge in the thoracic drain and sputum showing *Aspergillus fumigatus*. A thoracoscopic pleural decortication was performed at day 46. During this intervention the persistent esophageal perforation was sutured and covered by diaphragmatic flap. The evolution was then favorable and he was discharged at day 55, and continued antibiotics until day 64. The follow-up EGD at three months and at one year showed no recurrence of perforation.

DISCUSSION

The clinical presentation of the Boerhaave syndrome depends on the size of the esophageal breach, the localization, and the delay between the perforation and the diagnosis [9]. The Mackler triad is found in 25% of cases which are chronologically repeated vomiting, chest pain, and subcutaneous emphysema [10]. The chest pain is the constant element of this disease, which is progressive and generally not relieved by pain killers. The subcutaneous emphysema is one of the signs that can make suspect a pneumomediastinum. In case of diagnosis delay, symptoms and signs of sepsis or septic shock could appear often linked with associated mediastinitis [11].

Beyond a well conducted clinical examination, CT scan is currently the gold standard allowing not only to localize the injured esophagus segment, but also the potential complications as the pneumomediastinum, pleural effusion, and pneumothorax [12, 13].

During the initial medical care, it is important to correct if present the multiple organ failure and to prevent infection (mediastinitis) due to oropharyngeal and digestive germs diffusion, as well as fungus. It is thus recommended to early introduce two empiric antibiotics for a minimum of two months. This has to include a broad-spectrum antibiotic associated to a beta-lactamine with a beta-lactamase inhibitor, and also an antimycotic drug to avoid Aspergillus infection [14].

The EGD is currently under discussion during the acute phase due to the risk of perforation worsening despite 100% sensitivity and 93% specificity. The treatment of this affection is based on two pillars (medical and surgery), and its effectiveness depends on the early diagnosis [15, 16]. In case of early diagnosis and sepsis, surgery is preferred, whereas endoscopic approach is favored if the disease is diagnosed within 48 hours [17]. According to a recent review, endoscopic technics seems

to have good outcomes if performed within 24 hours in selected patient with stable vital signs [18]. Surgery is the key element for the patient recovery. Chest tubes prevent pleural effusion and pneumothorax which can aggravate the symptomatology. Even though endoscopy is controversial, esophagus endoprosthesis limits bacterial diffusion, clogs the underlying perforations, and avoids/postpones the thoracic surgery which is often too severe during the acute phase or even lethal in hemodynamically unstable patients [19–21].

Finally, it is essential to take into account that the patients suffering from this complex disease need a technical facility such as a university hospital and must be transferred within the 24 hours allowing a survival rate of around 60–70% [22].

CONCLUSION

The spontaneous esophagus rupture is nowadays a rare pathology with a poor prognosis due to its atypical clinical presentation making its diagnosis very challenging. The care is based on the early detection using CT scan and early administration of antibiotics covering oropharyngeal and digestive germs. The prognosis depends on the prevention of multiple organ failure and early access of surgery and endoscopy which are the reference treatments, generally in university hospital. Despite its scarcity the Boerhaave syndrome represents a differential diagnosis of chest pain which should be considered.

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Author Contributions

Christelle Margot – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Jérémy Desmercieres – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Guarantor of Submission

The corresponding author is the guarantor of submission.

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Consent Statement

Written informed consent was obtained from the patient for publication of this article.

Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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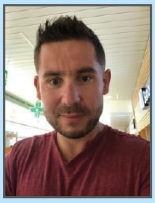

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