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Case report

Two-staged surgical management for complicated Boerhaave syndrome with esophagectomy and deferred gastroplasty: A case report

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ABSTRACT

Introduction and importance: Boerhaave syndrome is a rare, challenging entity with high morbimortality rates. Therefore, early diagnosis and prompt treatment are needed. However, a standardized technique has not been developed, especially in large esophageal ruptures.

Presentation of case: A female patient of 69 years with an acute thoracic syndrome consistent with severe retrosternal pain of sudden onset, radiating to the left hemithorax, vomiting, and dyspnea that began after food intake associated with subcutaneous emphysema, hypotension, and tachycardia. An A-CT was performed, revealing an esophageal perforation, and Boerhaave syndrome was diagnosed. The patient was taken to esophagectomy and gastroplasty. 2,5 years after the procedure, the patient was without long-term complications, and only dysphagia was present.

Clinical discussion: The differential diagnoses of acute thoracic syndromes are needed to be ruled out; however, it usually delays the diagnosis of Boerhaave syndrome. Therefore, early diagnosis (<24 h) may impact this patient's outcomes. On the other hand, esophagectomy can be feasible to control the acute condition and permit a digestive tract reconstruction.

Conclusion: In patients with large esophageal ruptures and concomitant septic shock, an esophagectomy is an option to control the source of infection and to permit early digestive tract reconstruction.

1. Introduction

Boerhaave's syndrome is the transmural perforation of the esophagus due a sudden increase in intraesophageal pressure [1], which can occur in any esophageal segment. It is a rare condition with high mortality rates [2]; therefore, it is essential to prompt diagnosis and treatment, which can vary according to the extent of perforation, the degree of necrosis of the esophageal wall and the clinical status [3]. The optimal therapeutic approach to this condition has not been developed, but surgery is the cornerstone.

We present the case of successfully surgical management of complicated Boerhaave syndrome in an academic center in Bogotá - Colombia, achieved with two spaced interventions: Primary esophagectomy and gastroplasty three days after. This work is in line with the SCARE 2020 criteria [4].

2. Case presentation

A 69-year-old woman with no previous medical history presented with severe retrosternal pain of sudden onset, radiating to the left hemithorax, vomiting, and dyspnea that began after food intake. She visited another hospital with these symptoms, where chest radiography showed a left pleural effusion. She was then transferred to our hospital after three days of onset of symptoms. On admission, she was conscious and oriented, with a heart rate of 117 bpm, blood pressure of 91/48 mmHg, respiratory rate of 24 rpm, a temperature of 37.5 °C, and oxygen saturation of 85 % with FIO2 at 50 %. Physical examination revealed subcutaneous emphysema in the right lateral region of the neck and decreased breath sounds with bibasal rales. The arterial blood gasses evaluation revealed pH 7.29, pO2 52 mmHg, pCO2 42 mmHg, HCO3–20 mEq/L, and lactate of 4.22. Hematological investigations revealed neutrophilia and elevation of acute-phase reactants. A chest X-

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ray in the emergency room showed massive left hydropneumothorax, initially treated with closed thoracostomy with adequate radiological control (Fig. 1A). Due to the persistence of symptoms, the emergency room team suspected acute thoracic syndrome, for which a CTA was performed, where findings suggestive of esophageal perforation were found (Fig. 1B), which was confirmed by an upper gastrointestinal endoscopy, which revealed a subcarinal 7 cm esophageal perforation a few centimeters below the aortic arch (Fig. 1C). Then, the thoracic team was consulted, and we diagnosed spontaneous esophageal rupture, i.e., Boerhaave's syndrome and septic shock requiring vasopressors and broad-spectrum antibiotics. Therefore, the patient underwent an exploratory surgery performed by teams of the thoracic and gastrointestinal surgery, where we found a right cloudy serous pleural effusion, complete esophageal dilatation, and an esophageal laceration in the middle 1/3 of the esophagus (immediately below the main carina) of 7 cm in length with significant perilesional necrosis respecting the lower 1/3 of the esophagus. Due to these intraoperative findings, both services decided that an esophagectomy for infection foci control with mediastinal exploration was required. We decided on a two-stage surgery due to the septic status secondary to infectious mediastinitis.

First, an esophagectomy to control the source of infection was performed by right postero-lateral thoracotomy access with preservation of the serratus muscle at the sixth intercostal space. The anterior and posterior mediastinal pleura were opened with the section of the azygos arch, and dissection of the middle 1/3 of the esophagus. Esophagectomy of the unhealthy wall was performed. The mediastinum's exploration, lavage, and drainage were performed finally with no acute complications. Parenteral nutrition was started.

The patient went to the ICU to control the infectious mediastinitis and septic shock. On the third postoperative day and once clinical improvement was evidenced, both surgery teams reconstructed the digestive tract with a transhiatal gastroplasty and mechanical endothoracic esophagogastric anastomosis with manual reinforcement by the Mikulicz type pyloroplasty. The neo-formed gastric tube's ascension to the mediastinum and right hemithorax was achieved (Fig. 2). No immediate complications were reported. In the same surgical procedure, left video thoracoscopy was performed to complete adequate decortication and lavage of the hemithorax. After seven days, endoscopic control was performed due to increased systemic inflammatory response, suspecting an anastomotic leak or a fistula formation; however, there were no early complications. In the third postoperative week, partial dehiscence of the anterior wall of the anastomosis was evidenced and treated with a self-expanding esophageal stent.

The patient stayed in the ICU to treat other medical complications of the infection. Parenteral nutrition was weaned progressively as the patient tolerated enteral nutrition. She subsequently presented pneumonia associated with mechanical ventilation, obstructive atelectasis due to mucus plug (treated with fiber bronchoscopy), pleural infection due to candida Albicans, prolonged intubation (requiring tracheostomy), sepsis, and bacteremia due to the peripherally inserted central catheter, and finally, renal failure treated with hemodialysis. After three months, the patient was discharged, and the esophageal stent was removed six weeks after placement with no evidence of leakage in the control esophagogram. At 2.5 years follow-up, the patient is under mild esophageal stenosis and dysphagia treated and controlled with endoscopic dilation with no further complications. The patient refers progressive improvement in quality of life.

3. Discussion

Boerhaave syndrome is a transmural spontaneous tear of the esophageal wall due to a sudden increase in intraesophageal pressure [5]. This rare clinical condition carries a high morbidity and mortality rate and is fatal in the absence of therapy [6]. In most cases, the perforations are usually longitudinal with a mean size of 22 mm and occur in the lower third of the esophagus due to a lack of anatomical support, an increased number of associated neurovascular structures, and a relative absence of longitudinal muscle fibers [7]. However, as seen in our case, the rupture can rarely occur in other locations, such as the middle third.

The classic Mackler's triad (vomiting, lower thoracic pain, and subcutaneous emphysema) is present in <50 % of cases [8], and due to the

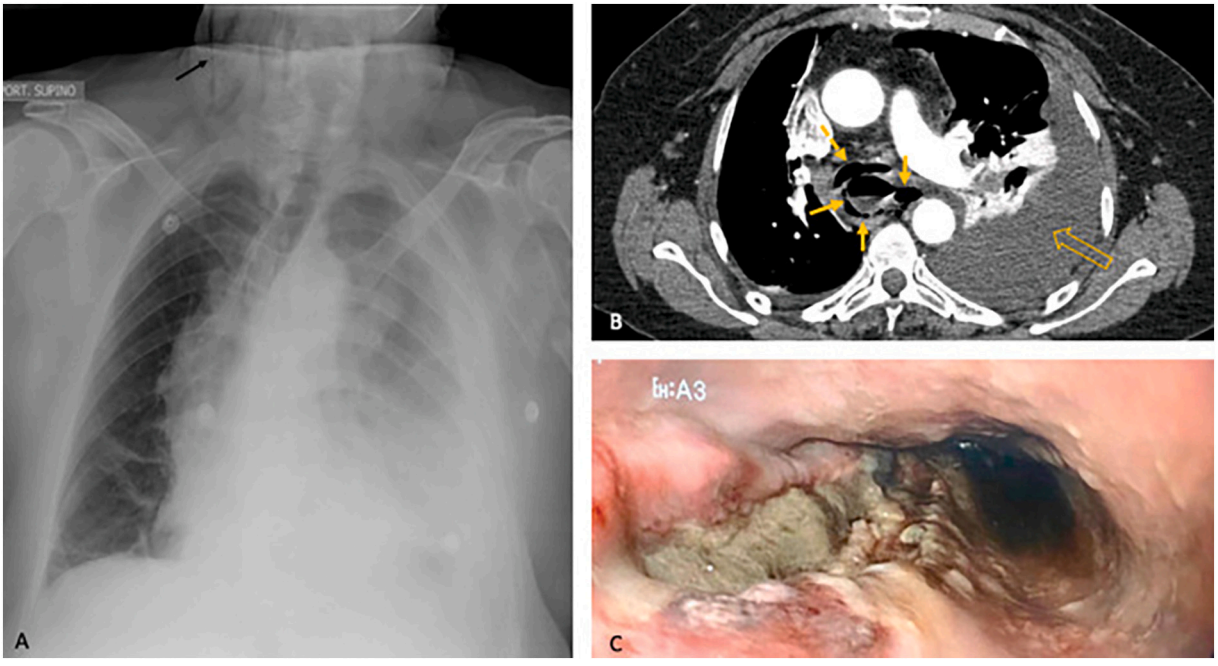

Fig. 1. Preoperative images of the patient. Image 1A shows a supine chest X-ray with evidence of left hydrothorax, where there is loss of the costophrenic angle and tracheal deviation to the right. Also there is subcutaneous emphysema shown by the black arrow. Image 1B is an axial cut of a preoperative computed tomography with intravenous contrast (A-CT) at the level of main bronchi emergence (dashed arrow) with evidence of pneumomediastinum (complete arrows) and a large pleural effusion (big arrow). Image 1C is an endoscopy showing the intraluminal defect in the esophagus in the left aspect of the image.

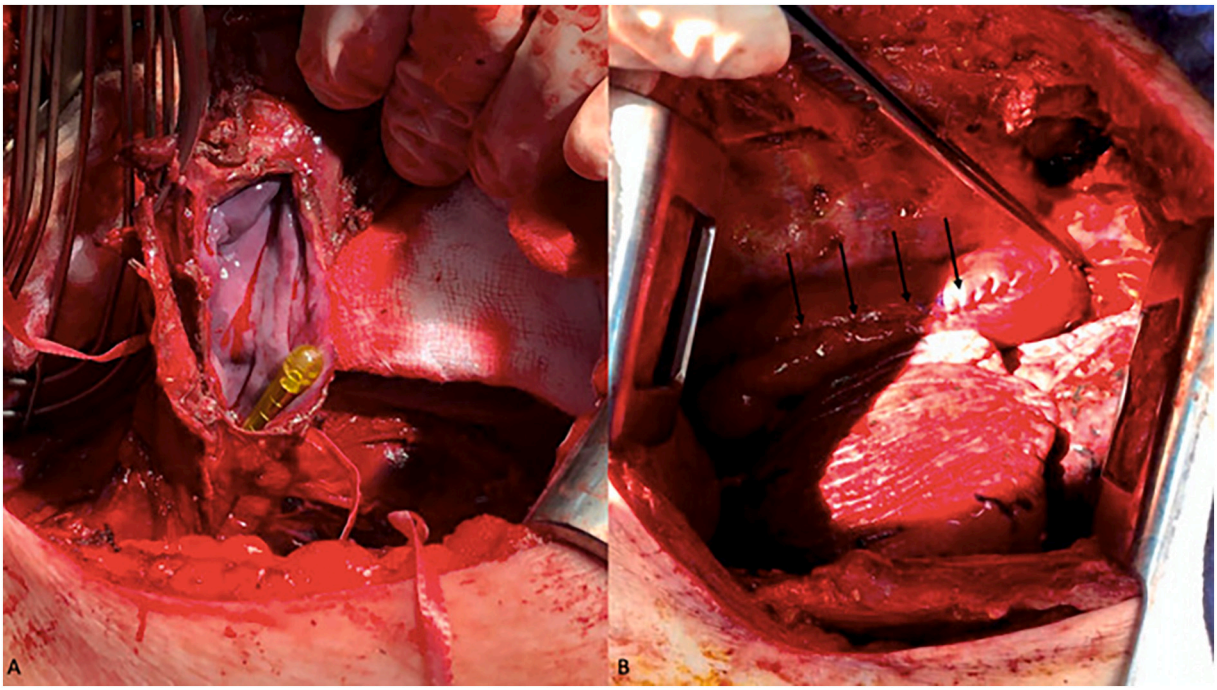

Fig. 2. Intraoperative images of the esophageal defect previous esophagectomy (Image 2A) through right thoracotomy with evidence of an irregular large defect of 7 cm with necrotic esophageal walls and gastroesophageal reconstruction with intrathoracic anastomosis (Image 2B).

nonspecific nature of symptoms, it may delay diagnosis and poor outcome. The initial prognosis of esophageal perforation will depend mainly on the appropriate treatment of mediastinal and pleural contamination [1,9], and the long-term survival will depend on the appropriate repair of the perforation. Consequently, the first objective of the surgical treatment must be the control of the infection so that later, and according to the patient's clinical conditions, definitive treatment can be carried out [5].

Dr. Herman Boerhaave first described the first case of Boerhaave syndrome in 1724 [10]; however, there is no consensus for optimal surgical management of this pathology. It depends on the location, severity, duration, degree of perforation, age, and patient condition. Schweigert et al. found that one of the most critical factors associated with mortality in patients with Boerhaave syndrome is the time between symptoms developed and the establishment of the diagnosis. They concluded that a diagnosis after 24 h was associated with higher mortality rates, with an odds ratio five times higher than those diagnosed before 24 h (OR, 4.65; 95 % CI, 0.33 to 265.80) [11].

Different therapeutic options have been described. Medical treatment is indicated for patients with minimal symptoms and when signs and symptoms of sepsis are absent (among others), which was not the case with our patient. Primary closure is feasible, especially in patients early diagnosed, but its performance lowers in late diagnosis [4,12]. Endoscopic stenting is a less invasive approach to treating an esophageal rupture. However, a retrospective study compared endoscopic stenting with operative management in patients with Boerhaave syndrome, and although they did not find differences in ventilation weaning, sepsis, or acute renal failure rates between both procedures, the endoscopic approach had higher re-intervention rates than surgical management, concluding that stenting alone was insufficient for the treatment of Boerhaave syndrome [11].

Esophagectomy is an invasive procedure indicated when the condition of the esophagus or the existence of underlying pathology suggests that primary closure of the perforation will not be possible [9]. The advantages and disadvantages of the procedure are summarized in Table 1 [13].

In our case, some aspects are important to highlight. Initially, the differential diagnoses of acute thoracic syndromes as a pulmonary embolism or an acute aortic syndrome are needed to be ruled out; however, it usually delays the diagnosis of Boerhaave syndrome. Our patient had the Mackler's triad, and appropriate diagnostic imaging was performed to rule out acute vascular syndromes and diagnose Boerhaave syndrome. The patient also arrived in septic shock with intraoperative findings reported, making the esophagectomy the best initial approach to control the acute condition and permit a digestive tract reconstruction in the same hospitalization. The patient's critical condition made it challenging to correctly examine the mediastinum with a minimally invasive approach. Although the esophageal course in the thorax can allow reaching the middle third through the left-side approach, we decided on a right thoracotomy approach because of better exposure of the middle 1/3 esophagus. Finally, we decided to do an intrathoracic gastroesophageal anastomosis to achieve less invasiveness due to performing a new procedure in the cervical area. Nutritional support was given through parenteral way and weaned progressively through the hospitalization.

Table 1.
Esophagectomy in Boerhaave syndrome [13].

Advantages	Disadvantages
Good control of septic source	Risk of dehiscence or strictures
Lower mortality rate versus conservative treatment	Increased operating time
Lower morbidity rate versus conservative treatment	
Lavage and drainage of mediastinum	

4. Conclusion

This case's primary interest is successfully treating an extensive esophageal perforation in a septic patient with a lousy prognosis through two sequential interventions: an initial surgery to control the infection (esophagectomy without reconstruction) and a second digestive system tract reconstruction surgery. We propose this approach as a feasible approach in severe cases with septic shock, large perforations with

necrotic borders, late diagnosis, and significant contamination of the mediastinum.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethical approval

Institutional Ethical Committee of Fundación Cardioinfantil-Instituto de Cardiología approved this case report (Ref Number: Acta 29-2022).

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Author contribution

Julián A. Ariza-Traslaviña: Conceptualization, Supervision, Investigation, Writing- Original Draft, Visualization, Validation and Project Administration. **Nicolás A. Caballero-Otálora**: Investigation and Writing- Original Draft **Camilo Polania-Sandoval:** Investigation and Writing- Original Draft **Carlos J. Pérez-Rivera**: Supervision, Writing – Reviewing and Editing, Validation Draft **Luis J Téllez-Rodríguez**: Supervision, Writing – Reviewing and Editing, Validation Draft **Manuel S. Mosquera-Paz**: Supervision, Writing – Reviewing and Editing, Validation and Project Administration

Guarantor

We designate Julián A. Ariza-Traslaviña as our guarantor, accepting full responsibility, assuring complete confidentiality of the data and that the study was conducted in a properly manner.

Research registration

N/A.

Declaration of competing interest

N/A.

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