Scholars Journal of Medical Case Reports
Abbreviated Key Title: Sch J Med Case Rep
ISSN 2347-9507 (Print) | ISSN 2347-6559 (Online)
Journal homepage: https://saspublishers.com

OPEN ACCESS

Gastroenterology

Mackler's Triad in Boerhave Syndrom: Exceptional Case Report

T. Adioui[1], S. Rokhsi[1*], F. Nejjari[1], S. Berrag[1], M. Tamzaourte[1]

[1]Department of Gastroenterology 1, Mohammed V Military Training Hospital, Rabat, Morocco

DOI: 10.36347/sjmcr.2022.v10i12.031 | **Received:** 18.11.2022 | **Accepted:** 27.12.2022 | **Published:** 30.12.2022

***Corresponding author:** S. Rokhsi
Department of Gastroenterology 1, Mohammed V Military Training Hospital, Rabat, Morocco

Abstract **Case Report**

Boerhaave syndrome is a rare entity, difficult to diagnose, with a potentially fatal outcome if not managed rapidly. We report the case of a female patient diagnosed throughout the triad of Meckler and confirmed with CT Scan using a water-soluble contrast product such as gastrografin showing pneumo-mediastinum with important subcutaneous emphysema. Clinically the patient was stable ruling out surgery but requiring a broad-spectrum intravenous antibiotic therapy with monitoring. The evolution was reassuring after a 15 day follow up. The barogenic nature of the rupture probably explains the wide mediastinal contamination, rapidly responsible for severe respiratory distress, sepsis and shock. A careful history and a detailed imaging examination allow a rapid diagnosis and therefore more caution treating the patient.

Keywords: Boerhaave's syndrom, Mackler triad, emergency, subcutaneous emphysem, Spontaneous pneumomediastinum.

INTRODUCTION

Spontaneous esophageal rupture, also known as Boerhaave Syndrome, referring to transmural rupture of the esophagus in healthy individuals, is a rare but potentially fatal condition.

The diagnosis of Boerhaave syndrome should not be missed or delayed, especially in some patients who present with symptoms leading to the misdiagnosis of other more common pathologies, such as acute myocardial infarction, acute pancreatitis or gastric ulcer perforation.

In this case report, we will detail the case of a patient presenting the pathognomonic triad of Mackler allowing us to establish an early diagnosis and thus avoid a less gloomy prognosis with a more conservative management.

CASE REPORT

A 23-year-old female patient with no specific pathological history who presented to the emergency department for puffiness of the left hemifacial side.

The detailed history reveals isolated episodes of severe vomiting for 72 hours, without any further details on their possible causes. The main complaints were a diffuse discomfort, dyspnea with chest pain, followed on the day of her admission by the appearance of edema of the left hemiface.

The initial clinical examination revealed a hemodynamically stable patient with a BP= 120/60 mmHg, a heart rate of 73 beats per minute and a respiratory polypnea of 20 cycles per minute.

The patient presented clinically with cervical and facial subcutaneous emphysema with edema of the left hemiface.

The biological workup showed hypokalemia at 3.1 mmol/l, an inflammatory syndrome with CRP at 25 mg/l, leukocytes at 16.3 G/l. Chest X-ray was considered normal. A thoraco-abdominal CT scan with ingestion of gastrografine revealed a pneumo-mediastinum of great abundance molding the diaphragmatic structures with an important subcutaneous emphysema dissecting the left jugal, bilateral basi-cervical, right axillary soft parts, partially extended backwards to the vertebral soft parts with individualization of air bubbles in intra canalary at the height of D4, D5 and D6. This allowed the final diagnosis of Boerhaave's syndrome.

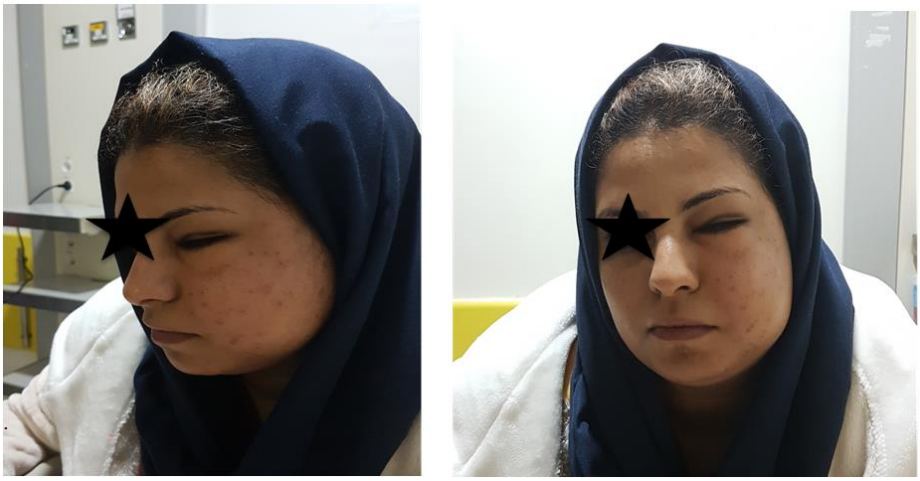

Fig 1: Picture showing the subcutaneous oedema of the left hemiface

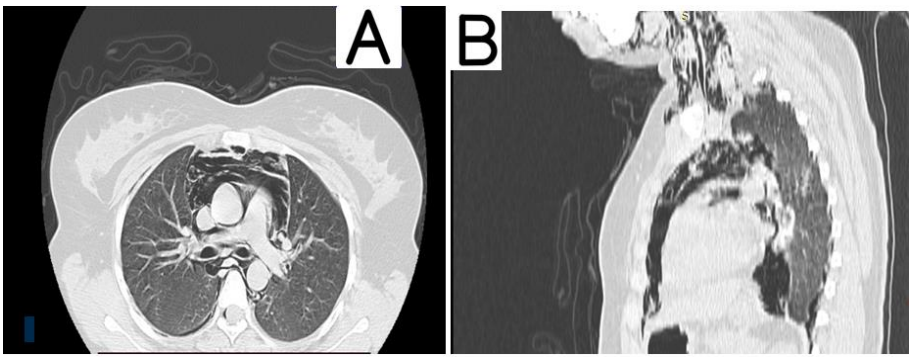

Fig 2: CT scan images showing the pneumomediastinum in axial (A) and sagittal (B) sections

A broad-spectrum intravenous antibiotic therapy was started. A surgical option was quickly ruled out given the patient's general condition and clinical stability.

The evolution was marked after 15 days of follow-up by the disappearance of the cutaneous emphysema.

DISCUSSION

First described by Hermann Boerhaave in 1724, spontaneous esophageal rupture (OSR) or Boerhaave syndrome, is a barotrauma due to a sudden increase in intraesophageal pressure (up to 200 mmHg), caused by intraluminal hyperpressure with a closed glottis [1]. The term "spontaneous" is used to distinguish Boerhaave's syndrome from iatrogenic, traumatic or tumor perforation esophageal ruptures.

Mackler's pathognomonic triad is rare on admission (vomiting, pain and subcutaneous emphysema) and is present in only 20% of cases [2], which is the case in our observation. Signs of acute respiratory distress¨ will rapidly dominate the clinical picture, testifying to pleural invasion.

In an emergency context, the history, the clinical examination, the electrocardiogram and the performance of non-invasive complementary examinations make it possible to eliminate the differential diagnoses of chest pain [3] more or less associated with a state of shock such as a cardiovascular origin (acute coronary syndrome, aortic dissection, pericarditis, pulmonary embolism), pleuropulmonary (pneumothorax, pneumopathy), digestive (biliary, pancreatic, gastric diseases), neurological, parietal, or functional diseases, and make the diagnosis of mediastinitis.

Radiological examinations are the only elements to reach the diagnosis. The later the workup, the more important the radiological findings. Although the chest X-ray is always abnormal (91% of cases), it is interpreted as compatible with an esophageal perforation only in 27% of cases [4]. During the first hours, the search for or discovery of pneumomediastinum is the main element of orientation [5].

In our case, a thoraco-abdominal CT scan confirmed the diagnosis of pneumomediastinum, which was initially suspected clinically in the presence of subcutaneous emphysema.

The thoracic CT scan therefore has a double interest in emergency: it allows a diagnosis of elimination (aortic dissection, fissuring of a thoracic

aortic aneurysm...) and directs the clinical investigation towards the etiologies of a pneumomediastinum. When this abnormality is observed, some authors have proposed to objectify the esophageal leak by absorption of gastrografine during the examination [6].

Digestive endoscopy is contraindicated because it is unreliable and traumatic [7, 3]. There is currently no specific consensus regarding surgical management in spontaneous esophageal ruptures, however, there is an undeniably predictable relationship between the time to diagnosis and initiation of appropriate treatment and patient prognosis.

Most centers where Booerhaave syndrome is treated use surgical techniques that include a variety of procedures including esophageal bypass, resection with delayed anastomosis, primary suture of the tear with mediastinal and/or pleural drainage, also performed thoraco- and laparoscopically [8]. Modern treatment methods involve endoscopic stenting, with or without pleural drainage, and clip placement [9, 10]. However, these are only one-time attempts and, based on the current state of knowledge, the effectiveness of this treatment is inconclusive.

All of the therapeutic approaches described above should be accompanied by modern antibiotic therapy and appropriate nutrition, either parenteral or intestinal via gastro or jejunostomy [8].

However, there are individual case reports of successful conservative treatment based on broad spectrum antibiotic therapy. The prerequisite for the use of non-invasive treatment in Boerhaave syndrome is a very early diagnosis (good general condition of the patient and absence of signs of infection) and excellent surveillance [10].

In our observation, the suggestive clinical presentation had allowed an early management and thus a most conservative treatment.

CONCLUSION

Boerhaave's syndrome is a rare and serious condition whose diagnosis remains difficult and the precocity of the management determines the prognosis of the patient.

An early surgical procedure may be avoidable when the clinical condition of the patient allows it, and leaves room for a less invasive and more conservative approach.

REFERENCES

1. Simonnet, B., Salaun, B., Thicoïpé, M., Jeannin, L., & Chanseau, P. (2009). Rupture spontanée de l'œsophage. *Journal européen des urgences*, *22*(1), 10-13.
2. Mutter, D., Evrard, S., Hemar, P., Keller, P., Schmidt, C., & Marescaux, J. (1993). Boerhaave syndrome or spontaneous rupture of the esophagus. *Journal de Chirurgie*, *130*(5), 231-236.
3. Ethuin, F., Marie, O., & Jacob, L. (1998). Médiastinites en dehors de la chirurgie cardiaque. Conférences d'actualisation. Paris: Elsevier; pp. 541-550.
4. Pate, J. W., Walker, W. A., Cole Jr, F. H., Owen, E. W., & Johnson, W. H. (1989). Spontaneous rupture of the esophagus: a 30-year experience. *The Annals of thoracic surgery*, *47*(5), 689-692.
5. Schmutz G. R., Chem R. K., & Massard, G. (1992). Ruptures et perforations oesophagiennes. Encycl. mèd. Chir. (Paris-France), Radiodiagnostic-Appareil digestif, 33090, A-15, 1-13.
6. Mikami, M., Nakamura, S., Koseki, T., Kanemura, T., Baba, M., Fujikawa, T., ... & Nagaoka, S. (1989). A Case of Esophageal Rupture Confirmed by Chest CT: Characteristic Changes of Chest Radiographs. *The Japanese journal of thoracic diseases*, *27*(5), 630-634.
7. Malledant, T., Tanguy, M., & Artus, M. (1986). Spontaneous rupture of the esophagus. *Ann Fr Anesth Reanim*, 5(2), 128-133.
8. Toelen, C., Hendrickx, L., & Van Hee, R. (2007). Laparoscopic treatment of Boerhaave's syndrome: a case report and review of the literature. *Acta Chirurgica Belgica*, *107*(4), 402-404.
9. Piardi, T., Petracca, M., Baiocchi, G. L., Tiberio, G. A. M., Marando, A., Coniglio, A., ... & Giulini, S. M. (2007). La sindrome di Boerhaave: esperienza personale orientata al trattamento chirurgico. *Annali Italiani di chirurgia*, *78*(3), 209-215.
10. Matsuda, A., Miyashita, M., Sasajima, K., Nomura, T., Makino, H., Matsutani, T., ... & Tajiri, T. (2006). Boerhaave syndrome treated conservatively following early endoscopic diagnosis: a case report. *Journal of Nippon Medical School*, *73*(6), 341-345.