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Presentation of Boerhaave's syndrome as an upper-esophageal perforation associated with a right-sided pleural effusion: A case report

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Abstract

BACKGROUND

Spontaneous esophageal rupture or Boerhaave's syndrome is a rare and acute disease with a high incidence of misdiagnosis and mortality. Here, we aimed to explore the clinical characteristics, diagnosis, treatment, and prognosis of spontaneous esophageal rupture, and also to analyze the causes of misdiagnosis during the treatment of spontaneous esophageal rupture.

CASE SUMMARY

The clinical features of the patient with spontaneous esophageal rupture misdiagnosed earlier as pleural effusion were retrospectively analyzed and the reasons for misdiagnosis have been discussed based on the current review of the literature. The patient was admitted to a local hospital due to shortness of breath accompanied by vomiting and abdominal distension for five hours. Based on the computed tomography (CT) scan analysis, clinically, right pleural effusion was diagnosed. However, the patient was unwilling to undergo a procedure for right closed thoracic drainage. The patient also had intermittent fevers against infection, and during the course of

treatment, he complained of chest pain, following which, he was transferred to our hospital. Liquid of grapefruit-like residue drainage was observed. The re-examination of the chest CT scans suggested the presence of spontaneous perforation in the upper left esophagus. Therefore, the patient underwent an urgent procedure of esophageal hiatus repair. Unfortunately, the patient died of infection and respiratory failure due to progressive dyspnea after surgery.

CONCLUSION

Spontaneous esophageal rupture is a rare disease associated with a high fatality. The patients don't present any typical clinical symptoms and the disease progress rapidly. This case report highlights the importance of a dynamic review of chest CT scan, not only for the initial identification of segmental injury but also for prioritizing subsequent treatment strategies. Moreover, we have presented some clues for clinicians to recognize and diagnose spontaneous esophageal rupture at rare sites (upper-esophageal segment) through this case report of spontaneous esophageal rupture that caused the patient's death. We have also summarized the reasons for the misdiagnosis and lessons learned.

Key Words: Spontaneous esophageal rupture; Chest computed tomography; Upper-esophageal perforation; Right-sided pleural effusion; Misdiagnosis; Case report

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Core Tip: Spontaneous esophageal rupture is a rare disease associated with a high fatality. We reported a case of spontaneous esophageal rupture misdiagnosed earlier as pleural effusion at an early stage and investigated the causes of its misdiagnosis, along with our experience during diagnosis and treatment. This case report also highlights the

importance of a dynamic chest computed tomography review, not only for initial identification of the injured segment but also for prioritizing subsequent treatment strategies. Moreover, we also provide clues for clinicians to recognize and diagnose spontaneous esophageal rupture at a rare site (upper-esophageal segment) by reporting this aforementioned case.

INTRODUCTION

Spontaneous esophageal rupture refers to the full-thickness rupture of the esophageal wall caused by indirect trauma, non-foreign bodies, non-esophageal, and/or adjacent organ disease; it is also known as Boerhaave syndrome[1]. While the incidence of this disease is low, it is easy to be misdiagnosed at an early stage and progresses rapidly. After the occurrence of an esophageal rupture, under the driving of negative pressure in the pleural cavity, stomach contents easily enter the mediastinum and thorax, which often causes serious mediastinum infection and empyema in the early stages. If not treated promptly, severe sepsis rapidly develops into multiple organ failure and even death, which is an emergency during thoracic surgery[2]. Therefore, the associated mortality rate is extremely high. Thus, correctly diagnosing spontaneous esophageal rupture in the early stage, is of great importance for the survival of patients with spontaneous esophageal rupture.

Herein, we reported a case of spontaneous esophageal rupture misdiagnosed earlier as pleural effusion at an early stage and investigated the causes of its misdiagnosis, along with our experience during diagnosis and treatment. Additionally, we also highlight the importance of reviewing dynamic chest computed tomography (CT) scans for the diagnosis of spontaneous esophageal rupture.

CASE PRESENTATION

Chief complaints

An 84-year-old male was admitted to a local hospital, with complaints of shortness of breath, abdominal distension, and vomiting.

History of present illness

An 84-year-old male was admitted to a local hospital, with complaints of shortness of breath, abdominal distension, and vomiting.

Additionally, the patient did not vomit again during his stay at the hospital.

Based on the evidence, the patient was diagnosed with pleural effusion and recommended to undergo a procedure for right closed thoracic drainage, however, the patient's family refused given his advanced age. Therefore, antibiotics were prescribed for preventing infection. However, after treatment, the blood inflammatory indicators were significantly elevated and did not improve [WBC: 19.40 × 10^9/L; C-reactive protein (CRP): 304.90 mg/L] (Table1). The patient also suffered from intermittent fevers and over time, complained of chest pain. After eight days, the patient was transferred to our hospital.

After admission, the patient agreed to undergo the procedure for right closed thoracic drainage and grapefruit-like residue drainage liquid was observed (Figure 1C). During the physical examination, subcutaneous emphysema of the right chest wall with crepitus was detected.

History of past illness

The patient had no history of lung diseases.

Personal and family history

No similar disease was found in his families.

Physical examination

His vitals were stable and no other specific signs were found.

Laboratory examinations

The initial blood routine examination results showed that both white blood cell (WBC) count and CRP levels were slightly elevated (WBC: 12.15 x 109/L, CRP: 13.96mg/L, Table1).

During hospitalization, laboratory tests of this patient also indicated an increase in the markers for inflammation.

Imaging examinations

The chest CT scan showed the presence of a small amount of fluid in the right pleural cavity (Figure1A). Thus, the chest CT scanning was repeated. Right-sided pleural effusion with right lung distension insufficiency and perforation of the upper left esophagus were observed (Figure 1B).

FINAL DIAGNOSIS

Considering the above signs and symptoms, the patient was diagnosed with spontaneous perforation of the upper left esophagus, and an urgent procedure for esophageal hiatus repair was performed.

TREATMENT

During surgery, the skin was incised after making a right lateral thoracic incision. The patient's right chest wall, muscles, and fascia were severely congested and edematous, along with a ruptured esophagus (Figure 1D).

OUTCOME AND FOLLOW-UP

Unfortunately, due to the deterioration of his conditions, the patient died from infection and respiratory failure.

DISCUSSION

Spontaneous esophageal rupture, a rare and life-threatening disease, was first reported by Rokicki M in 1724, and to date, a mere 50 cases have been reported in the literature.[3] Based on an epidemiological survey for this disease in Iceland, it has a low incidence of 31 per million per year[4]. Moreover, several studies confirm that men are more prone to morbidity than women and that the highest risk group included those in the age group of 40-60 years[5]. Spontaneous esophageal rupture caused by vomiting followed by a large meal often precipitates into secondary bacterial infections, which contribute to

50% of the total mortality[6]. Therefore, early diagnosis and surgical treatment are of importance for the treatment of this disease.

Although many cases of spontaneous oesophageal rupture have been reported, the lack of specific symptoms of this condition continues to pose a challenge[7]. Mackler's triad comprising an acute presentation of retching or vomiting, lower chest pain, and surgical emphysema, is a clinical manifestation with relatively high specificity for the diagnosis of the spontaneous esophageal rupture. However, its incidence is only about 14%[8,9]. Other signs, which are non-specific, including hemodynamic blood instability or the presence of the Hammer sign-on auscultation, can also help in assisting the diagnosis of the disease.[10] As secondary infection can irritate adjacent organs, symptoms including abdominal pain, nausea, chest tightness, shortness of breath, and dyspnea can also occur. Besides, elevated cardiac biomarkers and amylase also make it difficult to differentiate it from pericarditis, myocardial infarction, peptic ulcer, and other conditions. For patients with clinical suspicion of the disease, early chest CT examination is particularly important as it shows the manifestation of mediastinal or free peritoneal air as the first sign.

To the best of our knowledge, spontaneous esophageal rupture often occurs in the thoracic esophagus and its incidence in the upper thoracic esophagus is relatively rare. The reasons for this are broadly described as follows: the myometrium of the esophagus is divided into two, the inner ring and the outer longitudinal layer. Approximately 2 mm thick elastic fibers are sandwiched between the two layers. Owing to the lack of coherence in the anatomical structure of the esophagus, a sudden rise in intra-esophageal pressure (up to 290 mmHg) can lead to rupture at this altered anatomical structure of the esophagus[11]. While esophageal rupture occurs most commonly in the lower third of the left thoracic segment of the esophagus (80%), it is less frequent in the right esophagus, the upper thoracic, and ventral segments of the esophagus[12]. Among the physical signs, right pleural effusion is also uncommon. In the case of the upper thoracic esophageal perforation, prevertebral or subcutaneous air may be present[13]. Herein, we reported in detail, a case of a spontaneous esophageal rupture in the upper

thoracic esophageal perforation, with no obvious signs and symptoms in the early stage. Owing to the lack of a dynamic chest CT review early on, this disease was misdiagnosed.

Collectively, the reasons for the misdiagnosis are as follows: first, the on-admission chest CT report was only suggestive of a right-sided hydropneumothorax, inconsistent with CT presentation in most reports; additionally, the chest pains began later on during the course of disease progression, along with a lack of other typical manifestations. Finally, the upper thoracic esophageal perforation is a rare site of esophageal rupture and the dynamic chest CT scan was not reviewed during hospitalization, thereby leading to misdiagnosis early on and consequent delay in the appropriate treatment.

CONCLUSION

This case report highlights the importance of a dynamic chest CT review, not only for initial identification of the injured segment but also for prioritizing subsequent treatment strategies. Moreover, we also provide clues for clinicians to recognize and diagnose spontaneous esophageal rupture at a rare site (upper-esophageal segment) by reporting this aforementioned case of spontaneous esophageal rupture and summarizing the reasons for the misdiagnosis.

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