—Case Reports—

Boerhaave Syndrome Treated Conservatively Following Early Endoscopic Diagnosis: A Case Report

Akihisa Matsuda, Masao Miyashita, Koji Sasajima,
Tsutomu Nomura, Hiroshi Makino, Takeshi Matsutani,
Akira Katsuno, Junpei Sasaki and Takashi Tajiri

Surgery for Organ Function and Biological Regulation (Department of Surgery), Nippon Medical School Graduate School of Medicine

Abstract

We report on a 41-year-old man with hematemesis and severe epigastric pain. Three hours after the onset of symptoms, we carefully performed upper gastrointestinal endoscopy for suspected upper gastrointestinal perforation. Endoscopy revealed a 2.5-cm-long longitudinal laceration of the lower esophagus without active gastric or duodenal ulcers. The laceration could be partially closed with endoscopic clipping. Computed tomography of the chest revealed a small amount of extraluminal air in the mediastinum. Neither pleural effusion nor pneumothorax was detected. We treated the patient conservatively on the basis of the following factors: a stable general condition without sepsis, limitation of the esophageal disruption to the mediastinum, and early diagnosis. The treatment course was uneventful, and the patient was discharged from the hospital after we had confirmed with endoscopy that the esophageal ulcer was completely healed. Although Boerhaave syndrome is generally considered to have poor prognosis, conservative therapy may be effective in select cases with early detection of the perforation.
(J Nippon Med Sch 2006; 73: 341–345)

Key words: Boerhaave syndrome, esophageal rupture, conservative therapy, endoscopy

Introduction

Boerhaave syndrome, first reported in 1724 by Boerhaave[1], is a rare condition leading to a spontaneous esophageal rupture caused by forceful vomiting. Spontaneous esophageal rupture is often life-threatening and can cause severe mediastinitis and sepsis. It is generally believed that a major factor contributing to the poor prognosis is the difficulty of diagnosis. Initial symptoms, such as vomiting and epigastric pain, are generally nonspecific. Delayed diagnosis and treatment cause unfavorable outcomes.

Traditionally, immediate surgical treatment has been advocated for Boerhaave syndrome. However, in certain cases, nonoperative management, including broad-spectrum antibiotics and parental alimentation, has yielded good results[2–5]. We present a case of spontaneous esophageal rupture with early

Correspondence to Akihisa Matsuda, Surgery for Organ Function and Biological Regulation (Department of Surgery), Nippon Medical School Graduate School of Medicine, 1–1–5 Sendagi, Bunkyo-ku, Tokyo 113–8603, Japan
E-mail: a-matsu@nms.ac.jp
Journal Website (http://www.nms.ac.jp/jnms/)

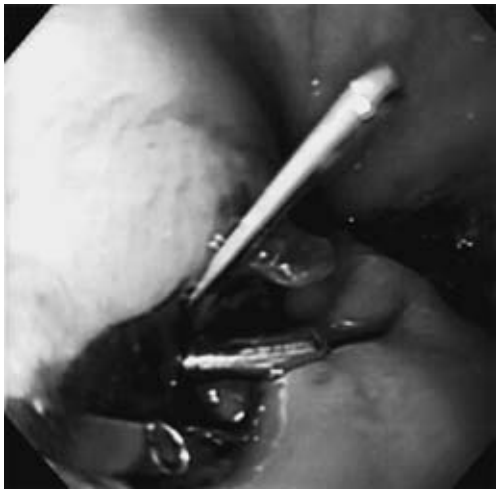

Fig. 1 Esophagoscopy at admission demonstrated a 2.5-cm-long longitudinal laceration of the lower esophagus. Endoscopic clipping was achieved only partial closure of the laceration.

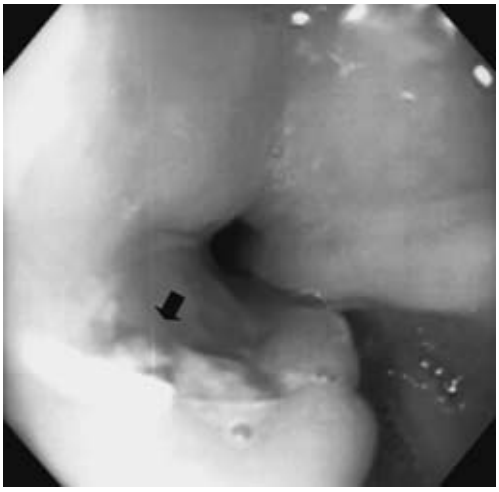

Fig. 3 Esophagoscopy on the 11th hospital day demonstrated a healing ulcer and an endoscopic clip **(arrow)** at the site of esophageal rupture.

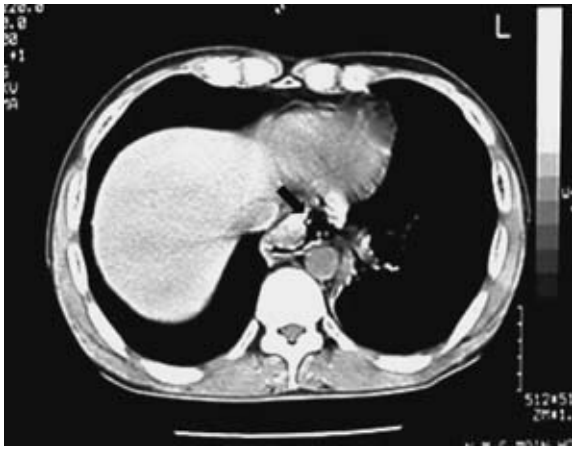

Fig. 2 Chest CT at admission demonstrated a small amount of extraluminal air in the mediastinum surrounding the lower esophagus **(arrow)**.

endoscopic diagnosis, in which a good result was obtained with conservative therapy.

Case Report

A 41-year-old man was referred to our hospital with hematemesis and severe epigastric pain in December 2002 (30 min after the onset of the symptoms). The patient had a past history of a duodenal ulcer at 21 years of age; however, he had no underlying esophageal disease. On admission, physical examination revealed severe abdominal tenderness with muscle rigidity in the epigastric region. The vital signs were as follows: body temperature, 39.0℃; blood pressure, 130/94 mmHg; pulse rate, 116 beats/min; and respiratory rate, 20 breaths/min. On auscultation, respiratory and heart sounds were normal. No subcutaneous emphysema was evident in the neck or upper chest region. The laboratory values on admission were as follows: white blood cell count, 11,500/μ*l*; red blood cell count, 308 × 10^4/μ*l*; hemoglobin, 11.6 g/d*l*; creatine phosphokinase, 674 IU/*l*; C-reactive protein (CRP), 4.69 mg/d*l*. Monocyte human leukocyte antigen (HLA)-DR expression, measured with flow cytometry, was 43.4%. A chest radiograph showed no pneumomediastinum and no free air in the subphrenic area. We initially suspected local peritonitis due to duodenal ulcer perforation. Three hours after the onset of the symptoms, we performed upper gastrointestinal endoscopy which revealed a 2.5-cm-long longitudinal laceration of the left posterior wall of the lower esophagus 38 cm from the incisors. Surrounding mucosal inflammation was comparably slight. No active gastric or duodenal ulcers were detected. Despite air infiltration into the esophagus, dilatation was insufficient (**Fig. 1**). With a diagnosis of spontaneous esophageal rupture, we attempted to close the laceration with endoscopic clipping. However, the laceration was too wide, and only partial closure was achieved. Computed

tomography (CT) of the chest revealed a small amount of extraluminal air in the mediastinum surrounding the lower esophagus. No pleural effusion or pneumothorax was demonstrated **(Fig. 2)**.

On the basis of several factors-a stable general condition without sepsis, limitation of the esophageal disruption to the mediastinum, and early diagnosis-conservative therapy was proposed with the provision that if the clinical status deteriorated, thoracotomy and drainage would be immediately performed. Immediately after admission, nasogastric suction was employed. The broad-spectrum antibiotic imipenem (1.5 g/day for 10 days), the proton-pump inhibitor omeprazole (40 mg/day for 10 days), and the protease inhibitor gabexate mesilate (2,000 mg/day for 5 days) were administered intravenously. Hyperalimentation was started on the second hospital day.

On the fifth hospital day, the patient was afebrile, and a follow-up CT scan of the chest revealed an absence of abscess formation and a significant decrease in extraluminal air in the mediastinum. Laboratory tests showed that the white blood cell count (7,900/μl) and the C-reactive protein level (0.01 mg/dl) had returned to within the normal ranges. Monocyte HLA-DR expression had recovered to 62.8% on the fourth hospital day and 85.2% on the ninth hospital day. On the 11th hospital day, esophagoscopy demonstrated a healing ulcer at the site of esophageal laceration **(Fig. 3)**, and esophagography showed no leakage of contrast agent (Gastrografin) from the esophagus. Therefore, oral intake was resumed. After we had confirmed with endoscopy that the esophageal ulcer was completely healed, the patient was discharged.

Discussion

The causes of esophageal rupture are various and are classified into three types: iatrogenic, traumatic, and spontaneous. Twenty percent to 40% of all cases of esophageal rupture are spontaneous [6,7]. Spontaneous esophageal rupture is extremely dangerous and occasionally fatal condition because it may rapidly progress to severe mediastinitis, sepsis, and multiple organ failure. A reason for the poor prognosis of spontaneous esophageal rupture is a delay in the diagnosis because of its rarity and lack of specific symptoms[7,8]. These symptoms are similar to those of perforated peptic ulcer, pancreatitis, myocardial infarction, dissecting aortic aneurysm, pneumonia, and spontaneous peumothorax[9,10]. We initially misdiagnosed the present case as a perforated duodenal ulcer, because of the patient's history. In fact, Kijima et al.[11] have reported that esophageal perforation was initially diagnosed in only 28% of 60 Japanese patients with spontaneous rupture.

In the diagnosis of esophageal rupture, esophagography is traditionally considered a safe and sensitive modality[12,13]. White et al.[14] have also reported that for cervical esophageal ruptures, esophagography is more sensitive than esophagoscopy; however, in thoracic cases, esophagoscopy and esophagography are equally sensitive. Kim-Deobald et al.[7] have demonstrated that misdiagnosis rate with esophagography is 36% and of particular note, only 50% of cases of acute rupture are correctly diagnosed. This lack of sensitivity may be due to tissue edema or muscular spasm. We consider esophagoscopy to be an extremely useful method for diagnosis because it allows direct vision of the esophageal rupture and evaluation for underlying esophageal diseases, such as cancer. Esophagoscopy can also be used to observe the healing course of an esophageal rupture, as in the present case. However, the insufflation of air into the thoracic space may increase the pressure of pneumothorax and cause the pneumomediastinum to worsen. Therefore, esophagoscopy should be performed carefully and quickly with minimal insufflation of air. CT is also an efficient diagnostic method for esophageal ruptures, because it can reveal pneumomediastinum, pneumothorax, and pleural effusion. White et al.[14] have reported that CT is a useful diagnostic method, particularly for patients without typical symptoms of esophageal rupture.

HLA-DR is a major histocompatibility complex (MHC) class II protein. MHC class II expression is important for antigen presentation to lymphocytes and for initiating the specific immune response[15].

After trauma, monocyte HLA-DR expression is decreased, a change that correlates with clinical outcome, particularly for infective complications[16]. In the present case, monocyte HLA-DR expression had decreased to 43.4% at admission, but recovered afterward. This favorable recovery of immunological competence supported the validity of conservative therapy for this patient.

The treatment of esophageal rupture has been discussed in many reports. Most reports state that surgical intervention is the optimal treatment[17,18]. Skinner et al.[17] have recommended surgical intervention for treating esophageal rupture regardless of its etiology. However, several reports suggest that in some cases conservative therapy is appropriate and achieves outcomes comparable to or better than those of surgical treatment[3–5,13]. Cameron et al.[3] have reported the following criteria for conservative therapy: (1) the esophageal disruption is confined to the mediastinum: (2) the cavity is well drained into the esophagus: and (3) the patient has minimal symptoms and no evidence of clinical sepsis. They have also reported mortality rates of 38% in patients treated surgically and of only 9% in patients treated conservatively. Shaffer et al.[4] have argued that the criteria of Cameron et al.[3] are too conservative and have suggested alternative guidelines for the selective use of nonoperative treatment: (1) clinically stable patients: (2) early rupture detection, before major contamination has occurred: and (3) esophageal disruptions are well contained within the mediastinum or a pleural loculus. However, Shaffer et al.[4] have recommended surgical treatment for most cases of Boerhaave syndrome because of the high possibility of contamination. Kataoka et al.[19] have reported that endoscopic clipping is an effective method for treating esophageal rupture because it is less invasive than other therapeutic methods and because closure of the rupture is confirmed by direct observation. In this case, we attempted endoscopic clipping but were only able to partially close the wide esophageal laceration. However, we speculate that the partial closure contributed to the control of the leakage from the esophagus to the mediastinum and prevented further enlargement of the rupture. Although the rupture in our patient was spontaneous, we selected conservative therapy on the basis of following factors: a stable general condition without sepsis, limitation of the esophageal disruption to the mediastinum, and early diagnosis. Additionally, we could confirm with endoscopy that little food residue was present in the stomach and we concluded that the possibility of contamination was slight. In fact, the thoracic cavity and the mediastinum were not contaminated.

Previously reported cases of spontaneous esophageal ruptures that were treated conservatively with the broad-spectrum antibiotic imipenem have had favorable outcomes[20,21]. Takishima et al.[22] reviewed five cases of spontaneous esophageal rupture and reported that bacterial cultures of the intrathoracic drainage fluid from all cases were positive with three cases having a mixed infection of Gram-positive and Gram-negative bacteria. This bacteriological result may support the validity of using the broad-spectrum antibiotics. It has been speculated that reflux and leakage of gastric acid and pancreatic protease into the thoracic cavity are involved in tissue necrosis and abscess formation. Therefore, using a proton-pump inhibitor and a protease inhibitor for patients with esophageal rupture may be reasonable.

In conclusion, first, it is important to bear in mind that patients who complain of epigastric pain or chest pain following vomiting may have Boerhaave syndrome. Second, careful esophagoscopy is extremely useful for diagnosis. Finally, in limited cases, including those diagnosed as Boerhaave syndrome, conservative therapy can be applied.

References

1. Boerhaave H: Atrocis, nec descripti prius, morbi historia. Secundum medicae artis leges conscripta. Lugduni Batavorum Boutesteniana, 1724. Translation by Derbes VJ, Mitchell RE. Bull Med Libr Ass 1955; 43: 217–240.
2. Ivey TD, Simonowitz DA, Dillard DH, Miller DW Jr: Boerhaave syndrome: successful conservative management in three patients with late presentation. Am J Surg 1981; 141: 531–533.
3. Cameron JL, Kieffer RF, Hendrix TR, Mehigan DG, Baker RR: Selective nonoperative management of contained intrathoracic esophageal disruptions. Ann

Thorac Surg 1979; 27: 404–408.
4. Shaffer HA Jr, Valenzuela G, Mittal RK: Esophageal perforation: a reassessment of the criteria for choosing medical or surgical therapy. Arch Intern Med 1992; 152: 757–761.
5. Lyons WS, Seremetis MG, deGuzman VC, Peabody JW Jr: Ruptures and perforations of the esophagus: the case for conservative supportive management. Ann Thorac Surg 1978; 25: 346–350.
6. Ohri SK, Liakakos TA, Pathi V, Townsend ER, Fountain SW: Primary repair of iatrogenic thoracic esophageal perforation and Boerhaave's syndrome. Ann Thorac Surg 1993; 55: 603–606.
7. Kim-Deobald J, Kozarek RA: Esophageal perforation: an 8-year review of a multispeciality clinic's experience. Am J Gastroenterol 1992; 87: 1112–1119.
8. Henderson JAM, Peloquin AJM: Boerhaave revisited: spontaneous esophageal perforation as a diagnostic masquerader. Am J Med 1989; 86: 559–567.
9. Nesbitt JC, Sawyers JL: Surgical management of esophageal perforation. Am Surg 1987; 53: 183–191.
10. Symbas PN, Hatcher CR, Harlaftis N: Spontaneous rupture of the esophagus. Ann Surg 1978; 187: 634–639.
11. Kijima M, Ito T, Nagao F: A review of 60 reported cases in Japan of spontaneous rupture of the esophagus including a report of author's case. Jikeikai Med J 1981; 28: 21–40.
12. Parkin GJS: The radiology of perforated oesophagus. Clin Radiol 1973; 24: 324–332.
13. Mizutani K, Makuuchi H, Tajima T, Mitomi T: The diagnosis and treatment of esophagal perforations resulting from nonmalignant causes. Surg Today 1997; 27: 793–800.
14. White RK, Morris DM: Diagnosis and management of esophageal perforations. Am Surg 1992; 58: 112–119.
15. Hershman MJ, Cheadle WG, Wellhausen SR, Davidson PF, Polk HC Jr: Monocyte HLA-DR antigen expression characterizes clinical outcome in the trauma patient. Br J Surg 1990; 77: 204–207.
16. Faist E, Mewes A, Strasser T, et al.: Alteration of monocyte function following major injury. Arch Surg 1988; 123: 287–292.
17. Skinner DB, Little AG, DeMeester TR: Management of esophageal perforation. Am J Surg 1980; 139: 760–764.
18. Goldstein LA, Thompson WR: Esophageal perforations: a 15 year experience. Am J Surg 1982; 143: 495–503.
19. Kataoka Y, Miyamoto H, Arai N, et al.: A case of iatrogenic instrumental perforation of esophagus healed by endoscopic clipping (in Japanese with English abstract). Kyukyu Igaku (Japan) 1996; 20: 743–745.
20. Sato T, Obinata I, Takahashi S, et al.: Spontaneous esophageal rupture successfully treated by conservative therapy: report of two cases. Surg Today 2002; 32: 421–425.
21. Nishimoto M, Ajiki T, Matsumori M, Koreeda T, Ajiki S: A patient with severe spontaneous esophageal rupture treated by conservative therapy (in Japanese with English abstract). Gastroentorol Endoscopy 2003; 45: 138–143.
22. Takishima T, Matsuzaki H, Mieno H, et al.: Spontaneous esophageal rupture—problem in diagnosis and therapy—(in Japanese with English abstract). J Jpn Assoc Acute Med 1994; 5: 663–672.

(Received, September 11, 2006)
(Accepted, October 25, 2006)