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A rare case of duodenal ulcer perforation accompanied by Boerhaave syndrome

Agata Dżeljilji, Wojciech Rokicki, Marek Rokicki

Department of Thoracic Surgery, School of Medicine with the Division of Dentistry in Zabrze, Medical University of Silesia in Katowice, Poland

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Abstract

Esophageal perforation is the fastest progressing and the most life-threatening disruption of gastrointestinal tract continuity. It must be regarded as an emergency condition that requires early diagnosis as well as very aggressive and rapid implementation of treatment in order to avoid serious complications and death. Methods of treatment for spontaneous esophageal perforation continue to be a matter of controversy. However, all authors emphasize that ultimate success depends largely on the time taken to establish the diagnosis. The authors of this study describe a rare case of duodenal ulcer perforation accompanied by Boerhaave syndrome.

Key words: duodenal ulcer perforation, esophageal perforation, Boerhaave syndrome.

Streszczenie

Perforacja przełyku to najszybciej postępujące i najbardziej zagrażające życiu przerwanie ciągłości przewodu pokarmowego. Należy je traktować jako stan nagły wymagający wczesnej diagnostyki i bardzo agresywnego, szybkiego leczenia w celu uniknięcia ciężkich powikłań oraz zgonu.

Metody leczenia samoistnego przedziurawienia przełyku do dzisiaj budzą wiele kontrowersji, jednak wszyscy autorzy podkreślają, że końcowy sukces zależy w głównej mierze od opóźnienia w ustaleniu rozpoznania. Autorzy opisują rzadki przypadek przedziurawienia wrzodu dwunastnicy z towarzyszącym zespołem Boerhaave'a.

Słowa kluczowe: przedziurawiony wrzód dwunastnicy, perforacja przełyku, zespół Boerhaave'a.

Introduction

One of the rarest forms of esophageal perforation is Boerhaave syndrome (BS). It is a spontaneous rupture of the esophagus caused by impaired coordination of the act of vomiting and attempts to stop it. Various scientific reports estimate it at 8-25% [1-3].

The classic symptoms of BS, described in 1952 and known as Mackler's triad, are observed in less than half of the affected patients; they include: severe and profuse vomiting, "acute sharp pain" behind the sternum and/or in the epigastrium, and the appearance of subcutaneous emphysema on the chest wall, neck, and face [2-9]. A combination of these symptoms may appear with diverse intensity (one of them may be dominant, e.g. strong substernal pain), which may consequently lead to diagnostic errors that often end in tragedy for the patient. This results primarily from delays in the establishment of proper diagnosis and introduction of surgical treatment (Table I).

Case study

The patient was a 59-year-old homeless man with alcohol dependence syndrome. On October 19, 2012, he was admitted to the Department of General Surgery of a regional Silesian hospital due to midepigastric pain which had been intensifying for the previous two days as well as dyspnea accompanied by dry cough. His medical history included one instance of profuse vomiting of gastric contents.

Examinations performed on admission revealed the presence of peritoneal signs involving the whole abdominal cavity as well as asymmetry of the vesicular murmur (R<L).

An abdominal X-ray examination was performed with the patient in a standing position. Subsequently, abdominal computed tomography (CT) was conducted (October 20, 2012), demonstrating the presence of free air under the dome of the diaphragm and fluid in the right pleural cavity (thickness: up to 18 mm).

After a short preparation, the patient was qualified for exploratory laparotomy, which revealed a duodenal perforation and ulceration of the gastric cardia. Due to the challenges associated with repairing the perforation, a gastric tube was introduced, and partial gastric resection was performed using the Hofmeister-Finsterer method. During the surgical procedure, a drop in the ventilation parameters was observed, and the vesicular murmur on the right side

Address for correspondence: Agata Dżeljilji, MD, Department of Thoracic Surgery, School of Medicine with the Division of Dentistry in Zabrze, Medical University of Silesia in Katowice, 13-15 3 Maja St., 41-800 Zabrze, Poland, phone: +48 668376010, e-mail: nose.army85@gmail.com

became less pronounced. Therefore, after closure of the abdominal cavity, double drainage of the right pleural cavity was introduced, aspirating 2000 ml of brown fluid. The patient's condition deteriorated systematically after the surgery. Chest CT performed on the 2nd postoperative day visualized an esophageal fistula, massive contrast leakage into the pleural cavity, mediastinum, and abdominal cavity along the wall of the gastric stump, and fluid in both pleural cavities.

Diagnosed with esophageal perforation, the patient was transferred to the Chair and Clinic of General and Thoracic Surgery in Zabrze for further treatment. After the performance of basic examinations, left-sided thoracotomy was conducted. The pleural cavity was opened, and 1400 ml of brown, cloudy fluid was aspirated. During the procedure, gangrenous changes were found in the mediastinum, extending to the level of the left pulmonary artery. The perforation was located immediately above the diaphragm, on the posterior wall of the esophagus; its size was estimated at 3 cm. Despite the fact that more than 48 h had passed since the development of the perforation, an attempt was made to perform primary surgical repair of the esophageal wall. Due to difficulties with accessing the injury site, the anterior wall was incised in order to uncover the perforation. The posterior wall was treated with single-layer repair, and the anterior wall was treated with double-layer repair. Three drains were introduced into the pleural cavity. After the procedure, the patient was mechanically ventilated, received broad-spectrum antibiotics, and was fed parenterally; on the second postoperative day, he underwent tracheotomy. Control examinations demonstrated a clear elevation of inflammatory markers: procalcitonin (PCT) 3.56 ng/ml, C-reactive protein (CRP) 331 mg/l. Two dye tests performed during the early postoperative period did not indicate the presence of an esophageal leak. On the 8th postoperative day, a follow-up chest CT showed contrast leakage from the lower segment of the esophagus, extending along the spine and into the right pleural cavity (Fig. 1). Therefore, a decision was made to implant an esophageal stent. After the procedure, the patient's condition stabilized, which allowed him to be extubated; on the 37th day of hospitalization, the tracheotomy tube was removed. Follow-up radiological examination showed no contrast leakage from the esophageal lumen. The follow-up laboratory investigation showed stabilization of the inflammatory parameters (PCT < 0.01 ng/ml). The patient was discharged in good condition, remaining under the supervision of the Thoracic Surgery Outpatient Unit. During this time, he fed normally, eating solid foods. In January, the patient was again admitted to the clinic in order to undergo removal of the esophageal stent using gastrostomy access. Several days after this procedure, signs of impeded esophageal passage appeared. Control X-ray of the upper gastrointestinal tract revealed an esophageal stricture (Fig. 2). Several attempts to widen the stricture resulted in only short-term improvement. The patient was qualified for a surgical procedure: the strictured segment of the esophagus (approximately 4 cm in length) was resected using a thick gastric tube, and an end-to-end anastomosis was performed. Directly from the operating theater, the patient was transferred to the intensive care unit (ICU), where he stayed for three days. The postoperative course was uneventful. Presently, the patient is fed orally, and his body mass has increased by several kilograms.

Tab. I. Boerhaave's syndrome – the most common diagnostic mistakes

Perforated peptic ulcer in the stomach or duodenum
Myocardial infarction
Pneumonitis
Acute pancreatitis
Dissecting aortic aneurysm
Pneumothorax
Pulmonary artery embolism
Appendicitis
Renal colic
Lung abscess
Mesenteric artery embolism
Pericarditis
Splenic hemorrhage
Incarcerated diaphragmatic hernia
Pyelonephritis

Discussion

Although the clinical signs of esophageal perforation have been described in a number of academic guidebooks and numerous scientific reports, diagnosing BS remains challenging in many cases. This stems from the topography of the esophagus, which passes through three different body regions (neck, chest, and abdomen); its perfora-

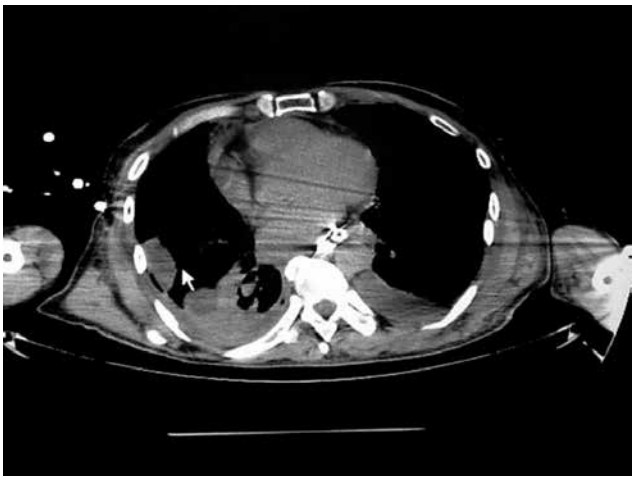

Fig. 1. Chest computed tomography with oral contrast (water solution). Visible fluid in both pleural cavities and contrast leakage outside the esophageal lumen

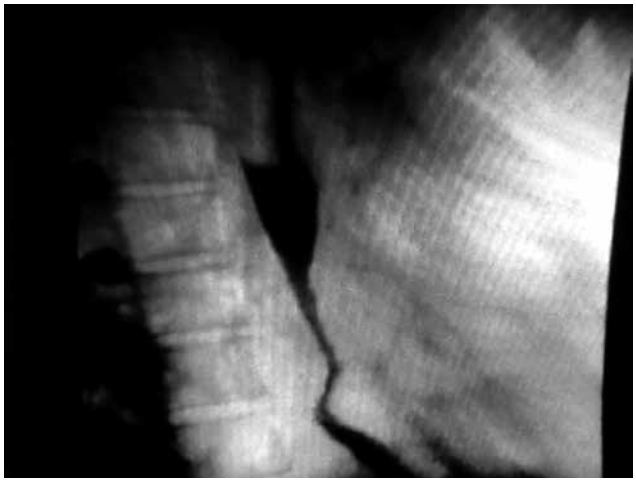

Fig. 2. X-ray of the upper gastrointestinal tract with contrast. Esophageal stricture

tion may, therefore, give diametrically different symptoms. The diagnostic difficulties are further compounded by the rarity of the disease.

At present, the most recommended method for diagnosing BS is CT using an oral contrast agent (water solution). Typically, the CT examination reveals the presence of air in the mediastinum and/or pleural cavity, esophageal wall injury, an esophagopleural fistula, pleural effusion, and the presence of a mediastinal abscess connected to the lumen of the esophagus. The sensitivity of this examination ranges between 92% and 100% [9-14]. Computed tomography is part of the standard diagnostic management in our clinic.

We did not perform an endoscopic examination (esophagoscopy) to diagnose Boerhaave syndrome as this method is not recommended by the literature as a first-line diagnostic tool (in spite of its 100% sensitivity and 83% specificity) because it is a risky procedure in patients in severe general condition. What is more, due to the necessity of air insufflation during esophagoscopy, there is a considerable risk of increasing the extent of the perforation by pumping air into the mediastinum and distributing the often limited purulent content across the whole pleural cavity. There are reports of patients in whom endoscopy of the upper gastrointestinal tract showed no esophageal perforation, and the presence of pathology was only confirmed after CT with contrast [14-16].

The primary factor affecting selection of the treatment method is the time from the moment of perforation until establishment of the diagnosis. Many authors believe that a delay in the start of treatment of more than 24 h increases the risk of death and complications by 50%, while primary surgical repair of the esophagus is associated with an over 20% risk of secondary leak development [17].

In the available literature, we have not encountered any reports describing concomitant ulceration of the duodenum and complicated BS. Acute abdominal signs with the presence of air under the diaphragm, observed by our colleagues from the regional hospital, clearly indicated the diagnosis of a perforated duodenal ulcer and largely "obscured" the signs of BS. It was only during the first procedure that the drop in ventilation parameters and the quieting of respiratory murmurs on the right side suggested the presence of pathology in the chest. The patient was brought to the clinic in a severe condition, with signs of sepsis. Despite the substantial delay in diagnosis (> 48 h) and the patient's condition after the partial resection of the stomach, an attempt was made to conduct primary repair of the perforated esophagus. Access to the perforation site (posterior wall, immediately above the diaphragm) was additionally impeded by the fact that we could not maneuver the esophagus due to concerns about tearing apart or damaging the already created gastrointestinal anastomosis – hence the decision to access the perforation site through the anterior wall of the esophagus. The secondary anastomotic leak observed on the 8th postoperative day was successfully repaired with a self-expanding stent. This technique is the recommended method for treating such complications [18].

The esophageal stricture after the removal of the stent was probably caused by the previously diagnosed ulceration of the cardia and, perhaps, the incision of the anterior wall. This could have consequently led to scarring and impeded esophagogastric passage. The aim of this report is to draw attention to the diagnostic and therapeutic challenges associated with BS.

Disclosure

Authors report no conflict of interest.

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