Spontaneous Esophageal Perforation: Atypical Presentation

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Abstract
A case of spontaneous esophageal perforation occurring in a healthy esophagus without any predisposing factor is reported. The problem of delayed diagnosis has been discussed.


Key words: Esophagus, rupture

Esophageal perforation may be traumatic or spontaneous, the former being commoner. Most spontaneous perforations are associated with a predisposing factor such as vomiting (Boerhaave's syndrome) or esophageal disease like carcinoma.\(^1\)\(^2\) We present a case of esophageal perforation with no history of vomiting and no evidence of esophageal disease at autopsy.

A 58-year-old non-alcoholic man presented with left-sided chest pain which had developed suddenly 3 weeks ago. He denied any vomiting or trauma preceding the onset of pain. Pain was persistent and associated with fever. He received antibiotics without any relief. He also developed jaundice two weeks after the onset of illness. Chest X-ray revealed left-sided pleural effusion for which an intercostal chest drainage tube was inserted and approximately two litres of pus drained. The patient was then referred to our hospital.

At admission, he was emaciated, toxic, febrile, anemic, jaundiced and dyspneic. His pulse rate was 110/min, blood pressure 130/70 mmHg, temperature 37.8°C and respiratory rate 24/min. Chest tube was draining small amount of thick pus. Chest X-ray revealed an air-fluid level in the mediastinum and haziness of left lung field. Methylene blue administered orally emerged from the chest tube, indicating an esophageal fistula. Gastroscopy study confirmed disruption of the lower esophagus with communication to the left pleural cavity. Investigations revealed: Hb 6.8 g/dL, TIBC 1400 μg/dL (neutrophils 80%), bilirubin 2.8 mg/dL, blood urea 125 mg/dL and serum creatinine 3.6 mg/dL. Blood culture was sterile.

The patient was resuscitated with intravenous fluids, blood transfusions, oxygen and antibiotics. Because of poor general condition, he was subjected to cervical esophagostomy, feeding gastrostomy and drainage of the left pleural cavity through two intercostal tube drains. He was electively ventilated in the postoperative period and antibiotics were continued. He, however, succumbed to septicemia on the sixth post-operative day. Autopsy revealed a 2 cm-sized perforation in the lower esophagus. Stomach was normal. A subpleural nodule, 1.5 cm in size, in the lingular lobe of the left lung revealed a granuloma without any acid-fast bacillus. No pathology was detected in the peri-esophageal lymph nodes or lungs apart from collapsed left lung.

Esophageal perforation is usually caused by external trauma, instrumentation or ingestion of foreign body or caustic agents, or is associated with esophageal diseases such as carcinoma, Barrett's esophagitis, rings or webs.\(^3\) Less than 20% of esophageal perforations occur in the absence of these and are designated as spontaneous perforations or Boerhaave's syndrome.\(^1\) Spontaneous perforations almost always result from increased intra-abdominal pressure transmitted to the esophagus against a closed glottis. Increased intra-abdominal pressure usually results from violent retching and vomiting and occasionally during defecation, parturition, seizures, weight-lifting or attack of bronchial asthma.\(^1\) The present case had no history of trauma, instrumentation or vomiting. A majority of the patients with spontaneous perforation do have episode of vomiting or other discrete events preceding other symptoms. A few cases of spontaneous perforation are known to occur in the absence of vomiting, but most of these ultimately prove to have underlying disease such as esophageal carcinoma-in-situ,\(^3\) peri-esophageal lymph nodal abscess,\(^4\) Absence of these could not be documented in the absence of surgical resection, autopsy or long term follow up.\(^5\)\(^6\) The present case had no esophagogastric or periesophageal disease at autopsy.

The classical triad of chest pain, vomiting and subcutaneous emphysema is rarely seen in patients with esophageal perforation and atypical presentation is well known.\(^5\) Subcutaneous emphysema is less frequently seen in thoracic esophageal perforation, which is the common site in patients with spontaneous perforation.\(^1\)

Early diagnosis and prompt treatment are critical in the management of esophageal rupture. Mortality is increased 2-4 times with a delay in diagnosis by more than 24 hours.\(^1\) Less than 50% of cases of spontaneous perforation are diagnosed within 24 hours, the initial diagnosis being pneumonia, lung abscess, pancreatitis, myocardial infarction or pericarditis.\(^7\) In the absence of vomiting as in the present case, a high index of suspicion is required to consider the diagnosis and investigate for the presence of esophageal rupture.
References


