Spontaneous esophageal rupture, report of 6 interesting cases

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Abstract:
Spontaneous esophageal rupture is a rare clinical condition, occurs most often in the lateral, lower 1/3 of the esophagus and is associated with a mortality rate of 20–75%. As a result of these nonspecific findings, Spontaneous esophageal perforation is often misdiagnosed as an aortic emergency, pericarditis, myocardial infarction, pulmonary embolus, spontaneous pneumothorax, perforated peptic ulcer, or pancreatitis. We outline 6 cases of spontaneous esophageal perforation came to our institution and will discuss about their diagnosis, required interventions, complications and 2-year follow up.

Keywords: Spontaneous esophageal rupture, case report, operation, follow up

INTRODUCTION
Spontaneous esophageal perforation is a rare clinical condition first described by the Dutch anatomist and physician Hermann Boerhaave in 1724. It represents 10-20% of esophageal ruptures and is defined as the spontaneous rupture of the total thickness of the esophageal wall
due to a barogenic trauma.[1-2] This occurs most often in the lateral, lower 1/3 of the esophagus and is associated with a mortality rate of 20–75% [3] Many patients present with symptoms such as chest pain, shock, or respiratory distress and physical exam findings are often nonspecific (tachycardia, tachypnea, or fever). As a result of these nonspecific findings, it is often misdiagnosed as an aortic emergency, pericarditis, myocardial infarction, pulmonary embolus, spontaneous pneumothorax, perforated peptic ulcer, or pancreatitis.Delayed diagnosis is one of the major differences in the management of iatrogenic esophageal rupture versus spontaneous rupture and may be responsible for the higher mortality rate in the latter [3] Here we present 6 cases of spontaneous esophageal rupture came to 5 Azar Hospital in Gorgan and will discuss about their diagnosis ,required interventions ,complications and follow up.

Case presentation:
During 2017 to 2020 we had 6 cases of spontaneous esophageal rupture. Only one patient was younger than 50 years old that had short segment esophageal necrosis because of food impaction. All others were older. Signs and symptoms consisted of chest pain, fever in favor of mediastinitis ,hydropneumothorax and empyema in chronic cases. Symptoms of food impaction were seen in one patient. Two patients had early diagnosis and four of them were diagnosed lately. In 2 cases the rupture was short segment and it was long segment in four others. Pathologic result was malignant only in the last case (6).
Based on clinical and radiological findings, appropriate intervention was determined. Surgical approach, morbidity (2-year follow up) and mortality are shown in below table.

<table>
<thead>
<tr>
<th>N</th>
<th>Age</th>
<th>Sex</th>
<th>Duration of rupture</th>
<th>Extent of rupture</th>
<th>Surgical approach</th>
<th>Mortality/morbidity</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>58</td>
<td>Female</td>
<td>Early diagnosis</td>
<td>Long segment</td>
<td>Right thoracotomy/esophagectomy/gastric pull up</td>
<td>posterolateral posterolateral posterolateral posterolateral posterolateral posterolateral</td>
</tr>
<tr>
<td>2</td>
<td>38</td>
<td>Male</td>
<td>Early diagnosis</td>
<td>Short segment</td>
<td>Right thoracotomy/distal esophagectomy/esophagogastric anastomosis</td>
<td>Mild reflux</td>
</tr>
<tr>
<td>3</td>
<td>65</td>
<td>Female</td>
<td>Late diagnosis</td>
<td>Short segment</td>
<td>Left thoracotomy/decortication/esophageal repair with diaphragmatic flap</td>
<td>-</td>
</tr>
<tr>
<td>4</td>
<td>75</td>
<td>Female</td>
<td>Late diagnosis</td>
<td>Long segment</td>
<td>Right thoracotomy/decortications/esophageal repair with pleural flap/stent indwelling was unsuccessful</td>
<td>Septic shock, death</td>
</tr>
</tbody>
</table>

Table 1- Six cases of Spontaneous esophageal rupture. Cerebrovascular accident (CVA)
<p>| | | | | | |</p>
<table>
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</thead>
<tbody>
<tr>
<td>5</td>
<td>85</td>
<td>Male</td>
<td>Late diagnosis</td>
<td>Long segment</td>
<td>Right posterolateral thoracotomy/decortications/esophagectomy/cervical esophagostomy and feeding jejunostomy/cervical reconstruction with gastric pullup 2 months later</td>
</tr>
<tr>
<td>6</td>
<td>75</td>
<td>Male</td>
<td>Late diagnosis</td>
<td>Long segment</td>
<td>Left posterolateral thoracotomy/decortications/esophagectomy/cervical esophagostomy and feeding jejunostomy/cervical reconstruction with gastric pullup 2 months later</td>
</tr>
</tbody>
</table>

Discussion:
The nonspecific physical exam findings and the lack of any classical symptoms of Spontaneous esophageal rupture often result in delayed and misdiagnosis of this rare and lethal form of noniatrogenic esophageal rupture. [3] Our patients symptoms were Mediastinitis, Empyema, Chest pain, pleural effusion, Pneumothorax, Fever, Dyspnea, respiratory symptoms, Mediastinal emphysema, Sepsis. Noniatrogenic, life-threatening esophageal rupture can occur in the absence of any preceding history of vomiting, seizure, or chronic cough in patients without underlying esophageal pathology.

The management of Spontaneous esophageal rupture, regardless of the specific cause, begins with cessation of oral intake, administration of intravenous fluids and broad-spectrum antibiotics followed by surgical or endoscopic treatment of the tear. An isolated nonoperative approach can only be taken in a minority of patients who have radiologic findings showing lack of mediastinal or pleural contamination and no systemic symptoms of infection at the time of presentation [1-3]. Traditionally, in esophageal perforation that was diagnosed late, diversion fistula is considered as the procedure of choice with the belief that reconstruction will be unsuccessful in a contaminated environment. As observed in our cases, primary repair is possible with low morbidity and mortality even in presence of delayed diagnosis and mediastinal abscess. Many therapeutic approaches are reported to treat spontaneous rupture of the esophagus, including usage of self-expandable covered metal stents and endo-clips. Although stents and endo-clips are becoming increasingly popular, this therapeutic option has limitations and the use should be considered carefully. [4-5]
Conclusion:
In conclusion, early diagnosis and repair decreases the mortality morbidity of patients with spontaneous esophageal rupture. There is still no established algorithm to guide endoscopic vs. surgical repair of esophageal perforation. Endoscopic closure should be attempted only in cases where there is absence of significant mediastinal contamination such as in early diagnosed iatrogenic rupture. Primary closure of late diagnosed Spontaneous esophageal rupture can be successful and it should always be attempted initially regardless of the time lapsed from rupture. It is critical to debride necrotic tissue, drain mediastinal space and close the esophageal defect to control continuous contamination to achieve success.

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