Recurrent spontaneous esophageal rupture

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Abstract

Spontaneous esophageal rupture is an uncommon and poorly understood condition. Recurrent rupture is extremely rare, with only one previously reported case in the literature. Here, we present a case series of two patients who had recurrent ruptures, and discuss the principles underlying the management of such cases.

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Keywords: Esophagus; Surgery; Rupture

1. Introduction

Although spontaneous rupture of the esophagus (Boerhaave’s syndrome) is a well-described condition, recurrent rupture is rarely reported. We discuss the management of two such cases.

2. Patient 1

A 59-year-old male presented with right-sided chest pain and a single episode of haematemesis. A water-soluble contrast swallow showed a spontaneous esophageal rupture and he underwent right thoracotomy for repair of a 3-cm tear in the distal esophagus. Following this, he made a smooth post-operative recovery; however, 6 weeks later, he noticed dysphagia to solids. Esophagoscopy revealed a smooth tight stricture at the level of the repair, which was dilated to 60F with a Maloney bougie. Post dilation, the patient’s swallowing remained satisfactory.

Twenty-six months later, he presented to the Emergency Department 1 hour following acute onset of right chest pain and haematemesis. He was dyspnoeic and hypotensive with a systolic blood pressure of 80. A chest radiograph showed a large right-sided pleural effusion, and a water-soluble swallow revealed an uncontained extravasation of contrast from the distal esophagus. In view of his early presentation and haemodynamic compromise, surgical repair was undertaken. A redo right thoracotomy was performed and a 2-cm tear in the distal esophagus at the site of the previous repair was identified. This was repaired in two layers and reinforced with a vascularised intercostal muscle flap. An esophageal biopsy was also taken, revealing Barretts metaplasia. The patient made an unremarkable recovery and on review, 10 months later, he has no dysphagia, regurgitation or heartburn.

3. Patient 2

A 49-year-old male presented with chest pain and a single episode of haematemesis. A water-soluble contrast swallow revealed an esophageal rupture and a left thoracotomy and repair of a 3-cm distal esophageal tear were performed. The patient was transferred to the Intensive Care Unit, and eventually made a complete recovery. At the time of discharge, the patient had no dysphagia, regurgitation or heartburn.

Twenty-seven months later he presented to the Emergency Department 3 hours after experiencing severe right-sided chest pain exacerbated by swallowing. The patient showed no evidence of sepsis and was normotensive. A chest radiograph revealed a small right-sided pleural effusion. An urgent water-soluble contrast swallow revealed a contained extravasation of contrast around the distal right lower esophagus. Esophagagogastroscopy confirmed the presence of 3 cm tear in the right lower esophageal wall. In view of the patient’s stable clinical condition, he was treated conservatively with nutritional support via a feeding jejunostomy. Repeat esophagagogastroscopy performed 27 days after admission revealed a diverticulum commencing 30 cm from the incisors; and Barretts metaplasia in the lower 5 cm of the esophagus. A contrast swallow confirmed the presence of a diverticulum with no evidence of leak. The patient experienced a complete recovery and as of 3 months following his second perforation, he has no dysphagia, regurgitation or heartburn.
4. Discussion

Despite advances in critical care management, spontaneous esophageal rupture remains a life-threatening condition. The key to successful management is early recognition, with most authors reporting improved outcomes in patients diagnosed within 18-24 h of rupture [1]. Although surgical repair is usually undertaken in such cases of early presentation, there is evidence that conservative management may be effective in cases of minimal contamination of the mediastinum and with minimal signs of sepsis [2].

In our two cases, we performed a thoracotomy on the patient with evidence of free leak into the thorax in order to evacuate the pleural cavity and repair the rupture. By contrast we adopted a conservative strategy in the patient with a contained leak and no evidence of sepsis. The fact that both patients had a satisfactory outcome illustrates that non-operative treatment is a viable option even for patients with early presentation of recurrent rupture.

The pathophysiological mechanisms underlying spontaneous esophageal perforation are poorly understood. Previous studies have suggested that inflammation and esophageal dysmotility are important predisposing factors [3,4] and it is interesting to note that both of our patients had metaplastic changes suggestive of chronic gastro-esophageal reflux. It is therefore tempting to speculate that the re-ruptures may have occurred as a result of ulceration at the level of the previous repair, secondary to underlying gastro-esophageal reflux disease. Whilst previous series have identified long-term complications such as reflux and dysphagia following spontaneous rupture [3,4], there have been no large-scale studies on this subject. These cases illustrate an unusual but important sequela of recurrent spontaneous perforation. This complication is rare—indeed, there has to date been only one previous report of recurrent spontaneous esophageal rupture [5].

In summary, we present two cases of recurrent spontaneous esophageal rupture. On the basis of our albeit limited experience, we believe this condition should be managed using the same principles that govern the treatment of primary spontaneous esophageal rupture.

References