Case report

Secondary infection of a pre-existing thoracic aortic aneurysm by iatrogenic oesophageal perforation with aorta-oesophageal fistula formation

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Abstract

We report a case of iatrogenic perforation of the oesophagus, with subsequent infection of a pre-existing thoracic aortic aneurysm and development of an aorto-oesophageal fistula resulting in life-threatening haemorrhage. This was successfully treated with endovascular aortic stent-graft using a Cook Zenith Tx2 device. Follow-up CT scan 3 months later showed that the aneurysm sac is getting smaller, and patient had no further septic or haemorrhagic complications. However, we emphasise that even if initial endovascular management of aorto-oesophageal fistula is successful, these patients often risk recurrent sepsis and therefore long-term clinical and radiological surveillance is mandatory.

Keywords: Iatrogenic oesophageal perforation; Thoracic aortic aneurysm; Aorto-oesophageal fistula; Endovascular aneurysm stent-graft

1. Introduction

The true incidence of iatrogenic oesophageal perforation following upper gastrointestinal (GI) endoscopy is variable, and depends on the experience of the operator, the disease process of the oesophagus, and whether any therapeutic procedures have been performed. Despite early diagnosis and treatment, this complication is often associated with significant morbidity and mortality, depending on patient’s medical condition, magnitude of underlying disease, and degree of contamination. Although open surgery remained the traditional management, published results on less invasive procedures such as oesophageal stents in treating iatrogenic perforation seem to be promising [1].

Similarly, the use of endovascular stent-grafts in treating mycotic aneurysms and aorto-enteric fistulae in the presence of sepsis is controversial. It may be a temporising measure in emergency situations, or sometimes the only option in the management of critically ill patients who are not medically fit for open surgery [2]. In the case of aorto-oesophageal fistula, although life-threatening haemorrhage will usual stop immediately with successful endovascular treatment, subsequent sepsis with recurrent haematemesis may be difficult to treat [3].

This is the first case in the literature reporting secondary infection of a pre-existing thoracic aneurysm caused by iatrogenic oesophageal perforation following an upper GI endoscopy. The mycotic aneurysm had subsequently eroded into the oesophagus and the patient presented with life-threatening haematemesis. This was managed successfully with endovascular repair, and follow-up CT scans show that the aneurysm sac is diminishing.

2. Case report

A 79-year-old man presented with progressively worsening dysphagia and hoarseness of voice. There was no history of haematemesis. CT scan showed a saccular thoracic aortic aneurysm with a maximum diameter of 5.5 cm (Fig. 1A). Upper GI endoscopy was performed showing external compression of the oesophagus by the aorta. However, he developed sudden onset severe chest pain and shock 1 day afterwards, and clinical diagnosis of iatrogenic oesophageal perforation was made. Emergency chest radiograph and CT confirmed pneumomediastinum (Fig. 1B). Gastrograffin swallow confirmed the perforation site at the level of the fifth thoracic spine, and the patient was transferred to our institution for further management.

The patient remained clinically stable after adequate oxygenation, fluid resuscitation, antibiotics therapy, and insertion of chest drain. He was treated conservatively with
total parental nutrition and a Polyflex® oesophageal stent was inserted to attempt to seal the perforation. Three weeks later, he developed sudden massive fresh blood haematemesis. Emergency upper GI endoscopy showed blood coming from behind the oesophageal stent (Fig. 1C). Clinical diagnosis of aorto-oesophageal fistula was made, and emergency endovascular stenting of thoracic aortic aneurysm was done using a Cook Zenith Tx2® stent-graft, with intentional covering of the left subclavian artery origin in order to provide an adequate proximal landing zone (Fig. 2A and B).

Patient recovered well after the procedure without further episodes of haematemesis, sepsis, or chest pain. Follow-up CT scan 3 months later showed successful shrinkage of the size of the aneurysm, and successful treatment of the aorto-oesophageal fistula (Fig. 2C and D). Although the patient did not have any positive blood or tissue culture, microbiologists had advised on long-term antibiotics in the forms of ciprofloxacin and amoxicillin/clavulanate.

3. Discussion

There are many different theories as to the aetiology of mycotic aneurysms. Most common of all is the result of systemic bacteremia and septic embolisation (such as in subacute bacterial endocarditis), which cause super-infection of a diseased and roughened atherosclerotic plaque acting as a culture media [2]. Rarely, organisms may colonise the intact vascular wall through the vasa vasorum, where the arterial wall is weakened by a local suppurrative process that results in aneurysm formation. A local extra-vascular infectuous focus may penetrate directly or via lymphatic tissue into the aorta, leading to necrosis of the wall, false aneurysm formation and subsequent rupture. Patients who are malnourished or immunosuppressed may be more prone to the development of mycotic aneurysms [2].

In rare cases, a pre-existing aortic aneurysm may become secondarily infected. Pre-existing aortic aneurysms which become secondarily infected, or septic patients who have an incidental finding of an aortic aneurysm can be differentiated from primary mycotic aneurysms by the fact that the aneurysms are not rapidly growing. This current case is the first in the world’s literature to report secondary infection of a pre-existing thoracic aneurysm by iatrogenic oesophageal perforation, with subsequent development of aorto-oesophageal fistula. The diagnosis of aorto-oesophageal fistula was made at upper GI endoscopy with blood coming from behind the oesophageal stent, and also gas in the aneurysm sac on CT.

Conventional open surgery on aorto-oesophageal fistula as a result of mycotic aneurysms carries high perioperative mortality. Even if the patient survives the initial operation, there is a life-long risk of aortic graft sepsis or rupture.
Whether endovascular repair is a temporising or permanent measure will often depend on the medical condition of the patient, and whether there is successful sealing and subsequent control of the sepsis and fistula. Despite isolated case reports and small clinical series to document success, there is no level 1 evidence to suggest that it will become standard treatment as long-term results are lacking. Topel et al. reported six patients with aorto-oesophageal fistula who were initially treated successfully with endovascular stent-grafts. Four patients subsequently underwent in situ repair of the descending thoracic aorta with cryopreserved homografts. With a mean follow-up period of 35 months, three patients have already died (including one who had subsequent open surgery), from septic or haemorrhagic complications [4]. Although postoperative antibiotic therapy is advocated as an essential adjunct to successful endovascular treatment of mycotic aneurysm, there are no definite guidelines on the optimal duration of antibiotic therapy [3,5]. Whilst most physicians would agree that antibiotics are required in the immediate postoperative period, the need for prolonged or life-long oral antibiotics is still debated and uncertain [5].

In time, there may be new methods of chemically binding antibiotics such as gentamicin to vascular prostheses covered with pig gelatine, which seems to be bacteriostatic to Escherichia coli, Pseudomonas aeruginosa and Staphylococcus aureus strains [6]. Graft material specifically able to resist microbes by either changing their surface characteristics or by incorporating antimicrobial substances within their structure is being developed, and these endovascular devices may be useful in treating aorto-oesophageal fistula.

Fig. 2. (A) Pre-stent-graft deployment showing the aneurysm sac. (B) Completion angiogram after successful deployment of the Cook Zenith Tx2 device with intentional covering of the origin of the left subclavian artery, with sealing of the aorto-oesophageal fistula. The oesophageal stent can also been seen clearly. (C and D) Three months follow-up CT scan with successful treatment of the aorto-oesophageal fistula. The aneurysm sac size was smaller, with no gas or collection detected in the sac or in the mediastinum. The oesophageal stent could be seen.

References