Unusual association of diseases/symptoms

Aorto-oesophageal fistula: an unusual complication of oesophageal biopsies

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Correspondence to Tom Wiggins, thomas.wiggins@hotmail.co.uk**Summary**

The authors report a case of an aorto-oesophageal fistula presenting in a patient undergoing investigation for possible oesophageal malignancy who underwent multiple oesophageal biopsies. A 73-year-old gentleman underwent endoscopic biopsy of an oesophageal lesion. Histology showed only inflammatory changes, endoscopy was repeated and again biopsy showed only inflammatory changes. The patient then presented with severe haematemesis, which was shown by CT angiogram to be due to an aorto-oesophageal fistula. This was treated with endovascular stenting, and the patient recovered well. Sequential imaging has shown gradual resolution of the submucosal swelling and lymphadenopathy, excluding the possibility of underlying oesophageal malignancy. This is the first reported case of aorto-oesophageal fistulation as a complication of endoscopic oesophageal biopsies. Aorto-oesophageal fistula is an uncommon but potentially life-threatening cause of haematemesis. It is a potential complication of endoscopic oesophageal biopsy.

BACKGROUND

Aorto-oesophageal fistula is a rare but potentially fatal cause for upper gastrointestinal bleeding. It was first described by Dubrueil in 1818.¹ We report a case of an aorto-oesophageal fistula presenting in a patient who was under investigation for possible oesophageal malignancy who underwent multiple oesophageal biopsies.

CASE PRESENTATION

A 73-year-old Caucasian gentleman underwent investigation for dysphagia at a District General Hospital. Endoscopy showed a 2-cm polypoidal lesion in the lower third of the oesophagus. This lesion was well-defined with no areas of necrosis (see figure 1). Initial central biopsies were taken using Boston Scientific Radial Jaw (Natick, Massachusetts, USA) 4 biopsy forceps. The biopsies revealed inflammatory changes alone without any evidence of dysplasia or neoplasia following which, a repeat endoscopy was planned. This showed appearances consistent with extrinsic compression or a submucosal lesion of the oesophagus. Deeper biopsies were taken at this stage which again showed inflammatory changes. CT scan suggested a diffuse soft tissue mass between the oesophagus and aorta at the level of the right atrium. A repeat endoscopy was further planned.

In the interim, the patient presented to the Emergency Department, with upper gastrointestinal haemorrhage. The bleeding was believed to be arising from a submucosal oesophageal tumour. Endoscopy at the local hospital was unhelpful due to the excessive bleeding. Following initial resuscitation, including a transfusion of four units, he was transferred to our tertiary referral centre for further assessment and definitive management.

On arrival, there was continued haematemesis. He was tachycardic and normotensive. His haemoglobin was 7.6 and a further three units of blood were transfused. While

still in the emergency department, the patient developed severe hypotension which was refractory to further fluid resuscitation. A CT angiogram was performed to identify the specific bleeding vessel with a view to embolisation.

INVESTIGATIONS

At CT, there was evidence of a pseudoaneurysm (14×11 mm) of the descending thoracic aorta with contrast extravasation outside the true lumen of the aorta extending into the oesophageal wall and lumen (figure 2). The overall appearances were consistent with an aorto-oesophageal fistula.

TREATMENT

Endoluminal stenting of the thoracic aorta was performed immediately for bleeding control. This was performed using two 26 mm×10 cm GORE TAG thoracic stentgrafts (W.L. Gore and Associates, Flagstaff, Arizona, USA). Following stenting, there was no evidence of further bleeding. The patient was treated with intravenous antibiotics, and was discharged after 7 days with long-term oral antibiotics.

OUTCOME AND FOLLOW-UP

Repeat CT scan after 6 weeks showed the thoracic aortic stent in good position with complete exclusion of the pseudoaneurysm (figure 3). The soft tissue thickening between the aorta and oesophagus had reduced in comparison to the previous imaging. No specific mass lesion was identified within the oesophagus. Repeat endoscopy continued to show extrinsic compression in the lower third of the oesophagus, but endoscopic ultrasound showed normal oesophageal wall layers, with some reactive lymphadenopathy present in the lower paraoesophageal area. Biopsies again showed inflammatory changes

Figure 1 Endoscopic appearances of lesion during initial endoscopy. Bottom right image represents lesion following biopsy.

Figure 2 CT showing area of pseudoaneurysm and contrast outside the lumen on the aorta delineating the aorto-oesophageal fistula.

alone. Sequential imaging has shown gradual resolution of the submucosal swelling and lymphadenopathy, excluding the possibility of underlying oesophageal malignancy.

DISCUSSION

The commonest causes of aorto-oesophageal fistula include thoracic aortic aneurysms (51% of cases) and foreign body ingestion (19%).² Other causes previously described include

Figure 3 Repeat CT scan showing aortic stent graft in position.

thoracic malignancies,³ oesophageal reflux,⁴ tuberculosis,⁵ caustic ingestion,⁶ congenital abnormalities,⁷ atherosclerotic disease⁸ and aorto-oesophageal fistula following traumatic injury.² Previously reported iatrogenic causes include a postoperative complication following open or stent graft repair of thoracic aortic aneurysms⁹ and postoesophagectomy.² Aorto-oesophageal fistula has also been described following oesophageal stent placement¹⁰ and nasogastric tube insertion.¹¹ It is the opinion of the authors that given the lack of any other underlying cause for aorto-oesophageal fistula in this case, endoscopic biopsy was the most likely precipitating factor for fistula development. This is the first reported case of aorto-oesophageal fistulisation as a complication of endoscopic oesophageal biopsies.

Presentation of aorto-oesophageal fistula is commonly a triad of symptoms, as described by Chiari,¹² that include mid-thoracic pain or dysphagia, sentinel minor haematemesis, followed by exsanguination after a short period.¹² The interval between initial haemorrhage and exsanguination is variable, and can often be more than 24 h.

The diagnosis of aorto-oesophageal fistula is commonly confirmed by CT angiogram. Although CT does not always demonstrate the fistula itself, it often delineates signs suggestive of its presence such as an aneurysm displacing the oesophagus and the presence of air bubbles within the thrombus.¹³ CT scanning can also show the extravasation of contrast outside the lumen of the aorta, as in our case.

Previously, the treatment for aorto-oesophageal fistulas was thoracotomy with aortic graft interposition, followed

by reconstruction of the oesophageal defect.¹⁴ Endovascular stenting is now increasingly used in the initial management of aorto-oesophageal fistulas for haemorrhage control. During endovascular stenting, graft material is being placed in a potentially infected area where oesophageal flora has contaminated surrounding tissues. As a result of this, stenting is sometimes used as a bridging technique until definitive open repair of the thoracic aorta and oesophagus is performed. However, in patients unlikely to tolerate open repair with no evidence of sepsis, endoluminal stenting with antibiotic cover can now be performed as a definitive procedure.

A series of six cases of aorto-oesophageal fistula, all treated primarily by endovascular stenting was described by Topel *et al.*¹⁵ Four of these cases were caused by thoracic aortic aneurysms, one case of arteria lusoria and one of unknown aetiology. Three of these cases required emergency stenting for bleeding control. Two patients were not candidates for later open repair due to their medical condition. Both these patients died within 8 weeks of discharge. One died from septic complications and the other from further gastrointestinal bleeding. The other four patients in this case series underwent subsequent open repair of the descending thoracic aorta. One patient died 1 year following treatment due to gastrointestinal bleeding. All other patients were alive at last follow-up (mean follow-up 35 months (range 2–76)). The management of aorto-oesophageal fistulas has been described in detail by Flores *et al.*¹⁶

Learning points

- ▶ Aorto-oesophageal fistula is an uncommon but potentially life-threatening cause of haematemesis. It is a potential complication of endoscopic oesophageal biopsy.
- ▶ Due to the severity of this condition, it is important to remain vigilant of this as a potential differential diagnosis for patients with severe upper gastrointestinal haemorrhage following upper gastrointestinal endoscopy.
- ▶ The technique of endovascular stenting for bleeding control appears to be a promising intervention for high-risk patients who are unlikely to tolerate an open repair. However, no long-term data for the efficacy of this treatment modality is currently available.

Competing interests None.

Patient consent Obtained.

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