CASE REPORT

Aorto-oesophageal fistula following TEVAR: an unusual cause of mediastinal air

M D Kay, B Davies, K Patel, D Gourevitch

University Hospitals
Birmingham, Birmingham, UK

Correspondence to
Mark Kay;
markuskayus@hotmail.com

SUMMARY

Aorto-oesophageal fistula (AEF) following thoracic endovascular aneurysm repair (TEVAR) has an incidence of 1.7%. Patients often present with constitutional symptoms or haematemesis and the condition is frequently fatal. We present a 79-year-old man who underwent TEVAR to exclude a ruptured 5.3 cm aneurysm. He re-presented with persistent fever and haematemesis and initially underwent oesophagoduodenoscopy and biopsy to exclude oesophageal carcinoma. Following CT scanning AEF was diagnosed and extra-anatomical reconstruction was performed. Two months later he suffered a sudden and unexpected large bleed into his ventilator circuits from his tracheostomy and died. Haematemesis following TEVAR should raise clinical suspicion of AEF. Early and accurate diagnosis is paramount in view of the high attendant mortality. CT should be considered the initial investigation of choice.

BACKGROUND

- Aneurysms are now most commonly managed by endovascular techniques and awareness needs to be raised that aorto-enteric fistula can occur following endovascular repair as well as open repair.
- If not suspected and treated early the outcome is poor.
- Gastrointestinal bleed is a common emergency presentation but this is an uncommon cause.

CASE PRESENTATION

A 79-year-old man was incidentally diagnosed with a 50 mm descending thoracic aortic aneurysm during preoperative assessment for coronary artery bypass graft. This was managed conservatively with annual surveillance CT imaging.

He presented acutely 3 years later with a ruptured 5.3 cm aneurysm rupture and underwent emergency endovascular stenting (Valiant Medtronic). A limited left thoracotomy was performed for evacuation of a haemothorax. Postprocedure imaging was satisfactory and the patient was discharged home.

Four months later he presented with cough, malaise and left-sided chest pain. CT angiogram demonstrated an endoleak (blood flow outside the stent filling the aortic sac) owing to a failure of the seal between the proximal graft fixation and the aorta (type 1 endoleak). This was managed by deployment of a further stent graft (Valiant Medtronic). Postoperative recovery was uneventful and he was discharged home 10 days later.

Two months later patient presented to his local hospital with anorexia, weight loss and

haematemesis and was investigated for a possible diagnosis of oesophageal carcinoma with an oesophagoduodenoscopy (OGD), biopsy and CT scanning.

INVESTIGATIONS

OGD suggested a 5 cm tumour beginning 30 cm from the incisors and biopsies were taken. These later revealed necrotic tissue only.

CT angiogram demonstrated para-aortic soft tissue shadowing and multiple gas locules surrounding the graft, suggesting AEF (figure 1). The patient was thus transferred to our hospital.

DIFFERENTIAL DIAGNOSIS

There are many potential causes of haematemesis such as oesophageal carcinoma or peptic ulcer disease, but in a patient with previous aortic surgery an AEF must be excluded before another diagnosis can be accepted.

TREATMENT

The AEF was managed by extra-anatomical reconstruction of the thoracic aorta (figure 2). A staged approach was utilised with a left carotid-subclavian bypass, followed by ascending–descending aortic bypass grafting and extirpation of the affected aorta. The oesophageal defect was closed with a double layer of 4/0 prolene and buttressed with omentum.

OUTCOME AND FOLLOW-UP

The patient had a stormy postoperative recovery in ITU with respiratory failure. Unfortunately 2 months later he suffered a sudden and unexpected large bleed into his ventilator circuits from his tracheostomy and died. Postmortem was unable

Figure 1 CT scan demonstrating mediastinal haematoma merging with perioesophageal fluid and gas locules surrounding the stent graft.

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Figure 2 Extra-anatomical reconstruction with ascending–descending bypass, reconstruction of the aortic arch and transaction of the aorta.

to identify the source of the bleeding but was felt to be due to a tracheo-inominate fistula.

DISCUSSION

AEF following TEVAR has a reported incidence of 1.7% which is comparable with open surgery.¹ Conservative management is normally always fatal. Open surgical and endovascular management have a 64% death rate.¹ Risk factors identified include having an emergency procedure, endoleak, pseudo-aneurysm or graft infection. This is thought to be owing to compression of surrounding structures and the local inflammatory response to such complications. Bacterial

colonisation of aortic thrombus (which is retained during endovascular stenting) has also been proposed as a source of graft infection.² This patient had several risk factors including an emergent procedure and a postoperative endoleak. The peri-graft haematoma cultured intraoperatively, grew *Hafnia alvei*, coagulase negative *Staphylococcus* and mixed anaerobes indicating graft infection.

The clinical presentation of AEF was originally described as a triad of mid-thoracic pain, sentinel arterial bleed (herald bleed) and exanguination after a brief interval.³ Constitutional symptoms of anorexia, fever and tiredness are also now known common presenting symptoms as witnessed in this patient. More than half of patients present with a herald bleed, which allows a small window of opportunity for diagnosis and planning of appropriate treatment. Other patients may present with heavy bleeding and haemodynamic compromise which is normally fatal.

Early diagnosis and management of this condition is thus paramount. There are many potential causes of haematemesis such as oesophageal carcinoma or peptic ulcer disease, but in a patient with previous aortic surgery an AEF must be excluded before another diagnosis can be accepted. The consequences of misdiagnosis, delayed diagnosis and its sequelae may prove fatal. Commonly employed diagnostic tools include OGD and cross-sectional imaging (CT), which have relative pros and cons.

OGD allows direct visualisation of the problem and is believed to be a sensitive and specific diagnostic study for AEF.⁴ It can also exclude other potential causes of bleeding. There are associated risks of endoscopic assessment which thus needs to be performed with care as dislodged clot during the procedure may result in uncontrolled bleeding. It requires a skilled endoscopist who can inspect down to the fourth part of the duodenum. Biopsies are contraindicated in cases of diagnostic doubt owing to the risk of bleeding. Endoscopy is also an invasive procedure for an already compromised patient that may require sedation. There can also be diagnostic delay if resources do not allow prompt examination. In this patient the diagnosis of AEF was not evident on OGD and biopsies were taken due to a suspected diagnosis of carcinoma.

Contrast-enhanced CT may demonstrate the fistula, or graft complications such as endoleak or infection revealing peri-graft gas/soft tissue shadowing and chest consolidation. CT is widely available, quick to perform and non-invasive. It can also be used to plan ongoing management. The risks of contrast-induced nephropathy needs to be balanced against the risk of diagnostic delay, but in the authors opinion should be the initial investigation of choice.

Other potential diagnostic tests include angiography and more recently a combination of fluorodeoxyglucose-positron emission tomography and CT.⁵ These diagnostic tools require skilled personnel to perform and interpret them and resources that are not available in many hospitals.

Although uncommon, aorto-enteric fistula must be considered in patients who had a TEVAR and presents with haematemesis or signs of sepsis. A high index of suspicion and early diagnosis are paramount, as the clinical course may be brief before fatal exanguination. Increased awareness among colleagues is vital. Repair should ideally be attempted in a centre cognisant with both open and endovascular aortic approaches to optimise the chances of a satisfactory result and recovery.

Learning points

- ▶ To suspect and investigate for aorto-enteric fistula in any patient who has had aortic surgery (endovascular or open) and presents with haematemesis or signs of sepsis.
- ▶ Increased awareness among colleagues is vital.
- ▶ A high index of suspicion and early diagnosis are paramount.
- ▶ Constitutional symptoms are common presenting features.
- ▶ Do not attempt to biopsy.

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