Aortoenteric fistula causing massive bleeding

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Abstract - Objective: Aortoenteric fistula (AEF) is a rare but potentially lethal condition. We present a typical case report and give a brief overview of diagnosis and treatment. Patient: An 83-year-old man presenting with recurrent haematemesis, melaena, and collapse four years after aortic surgery. Measurement and results: Gastroscopy showed a focal defect in the duodenum, and angiography confirmed the presence of an actively bleeding AEF. An endovascular graft was deployed (endovascular aortic repair, EVAR) and activated factor seven was administered, which controlled the bleeding. The patient subsequently died of multi-organ dysfunction syndrome. Conclusion: AEF is a life-threatening condition. A high index of suspicion, urgent imaging, and treatment are essential. EVAR can be used as a bridge to conventional surgical treatment, or as definitive treatment in patients not suitable for surgery. Activated factor VII might be used as an adjunct to achieve haemostasis.

Keywords - aortoenteric fistula, massive bleeding, activated factor seven, shock

Introduction
An aortoenteric fistula (AEF) is a rare but potentially life-threatening condition. In this case report, we describe a patient presenting with gastrointestinal bleeding and collapse. Endoscopic and radiological features as well as management of AEF are discussed.

Case report
An 83-year-old man presented to a small regional hospital with recurrent haematemesis, melaena, and collapse. His past medical history revealed open abdominal aortic aneurysm repair with a Dacron tube graft four years earlier. Other co-morbidities included hypertension, ischaemic heart disease, gastro-oesophageal reflux disease, and osteoarthritis. He was resuscitated and urgently referred to our centre. Because he was haemodynamically unstable, he underwent an urgent gastroscopy in the emergency department. This revealed fresh transported blood in the stomach and the duodenum. In the third part of the duodenum, a focal defect was noted with no active bleeding at that moment (Figure 1). This defect was suspected to be an AEF and the decision was made to attempt to stop the bleeding with an endoluminal aortic stent (endovascular aortic repair, EVAR). Subsequent angiography showed the fistula with active bleeding into the duodenum from the region of the distal anastomosis of the tube graft (Figure 2). A Cook Medical Zenith Endovascular Graft (Cook Australia Pty. Ltd.) was deployed successfully, covering both the proximal and distal anastomoses of the original tube graft (Figures 3A and 3B), and no further bleeding could be identified following deployment. Unfortunately, after arrival in the ICU the patient became haemodynamically unstable and the massive transfusion protocol had to be initiated. He received a total of 17 units of packed red blood cells, 16 units of fresh frozen plasma, and 5 mega-units of platelets. Despite application of the massive

Figure 1. Endoscopic image showing the duodenal lumen (white arrow) and a focal defect in the duodenum, suspected to be an aortoenteric fistula (AEF) (black arrow).
transfusion protocol, the haemoglobin level continued to drop and a significant coagulopathy persisted, which was indicative of uncontrolled bleeding. Activated factor VII (Novoseven, dose 90 mcg/kg intravenously) was administered, and the need for ongoing transfusion ceased. Following this, a repeat gastroscopy showed no active bleeding. A haemostatic clip was deployed over the duodenal defect to help prevent re-bleeding. Despite all our efforts, our patient developed progressive and severe multi-organ dysfunction syndrome. On day 4 following the EVAR, the decision was therefore made to withdraw treatment and he subsequently passed away. The coroner did not take jurisdiction.

Discussion

Aortoenteric fistula (AEF) formation is a devastating condition that can occur as a primary or secondary phenomenon (i.e. after previous aneurysm repair). Estimates of the incidence following aorta surgery range from 1% to 2%[1]. The complication is even less common after implantation of an aortic endograft [1,2]. With treatment, the 30-day mortality has been reported as 21% to 43%, with shock at presentation and the need for preoperative transfusion as poor prognostic indicators [3,4].

Prompt diagnosis of AEF is imperative for patient survival. Patients present with signs and symptoms of gastrointestinal bleeding and abdominal pain with or without signs of systemic infection and are often in a very poor clinical condition. However, AEF may also be clinically occult with a non-specific presentation, especially in primary AEF [5].

Because clinical signs may not be present or may not be sufficiently specific, imaging is necessary to achieve an accurate diagnosis. Although no single imaging modality demonstrates the condition with sufficient sensitivity and specificity, computed tomography (CT) has become the imaging modality of choice for evaluations in the emergency setting, owing to its widespread availability and high efficiency. CT has widely variable sensitivity (40%-90%) and specificity (33%-100%) for the diagnosis of AEF. Mimics of AEF include retroperitoneal fibrosis, infected aortic aneurysm, infectious aortitis, and peri-graft infection without fistulization. Differentiation is aided by the observation of ectopic gas, loss of the normal fat plane, extravasation of aortic contrast material into the enteric lumen, or leakage of enteric contrast material into the paraprosthetic space; these features are highly suggestive of AEF in a patient with bleeding in the gastrointestinal tract [6].

Conventional treatment consists of extensive open surgery (extra-anatomical bypass and aortic ligation), closure of the fistula tract, and complete removal of any prosthetic material. This treatment is associated with high morbidity and mortality, and therefore more minimally invasive options with endovascular repair have been attempted. Endovascular repair is often successful in the short-term, achieving favourable immediate outcome. In the presence of systemic infection, however, EVAR alone as an ultimate solution is often followed by repeat infection and bleeding [7]. A staged combination of EVAR treatment for acute bleeding and aggressive infection treatment with systemic and local antibiotics, surgical abscess revision, and fistula tract closure might be an option in fragile patients. For patients fit for open repair, EVAR can be used as a bridging procedure to definitive repair, particularly in the setting of systemic infection [8]. Recently, in a multi-centre retrospective comparative study of 25 patients with AEF, the EVAR as compared with an open repair resulted in lower immediate postoperative morbidity and mortality, with similar overall long-term survival rates [9]. Comparison of different surgical techniques in another retrospective cohort study reveals that in situ reconstruction and extra-anatomic reconstruction (e.g. axillofemoral bypass grafting) have comparable short- and long-term results, with reinfection rates after one and two years of 24% and 41% respectively [3].

To our knowledge, the use of activated factor VII to aid in haemostatic control of AEF has not been described before.

Conclusion

AEF is a life-threatening condition. A high index of suspicion, urgent imaging, and treatment are essential. EVAR can be used as a bridge to conventional surgical treatment, or as definitive treatment in patients not suitable for surgery. Activated factor VII may be considered as an adjunct to achieve haemostasis.

Figure 2. Coronal angiographic image during positioning of the stent graft with contrast leaking into the duodenal loop (black arrows) from the region of the distal anastomosis of the previous surgical graft.
Figure 3. Contrast-enhanced CT scan with A) an oblique curved reformat through the aorta and right iliac artery, and B) an oblique axial reformat through the distal aorta post-endovascular aortic repair (EVAR) to show the duodenal loop intimately related to the distal abdominal aorta (white arrow) and the stent graft passing through this region.

References