CASE REPORT

Secondary aorto-enteric fistula presenting over a 2-month period with recurrent gastrointestinal bleeding

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SUMMARY

A female in her sixties presented with recurrent gastrointestinal bleeding over a two-month period requiring multiple hospital admissions. Inconclusive upper and lower gastrointestinal endoscopies in addition to an initial negative CT angiogram initially left the diagnosis uncertain. A subsequent catastrophic bleed prompted a further CT angiogram that demonstrated a fistula between the aorta and third part of the duodenum with active contrast extravasation. Emergency surgery was carried out but ultimately the patient did not survive.

BACKGROUND

This phenomenon, although rare has been the subject of ongoing research and deliberation, most likely linked to its high mortality. A recent literature review looked at 332 individual cases and 1135 patients from previous papers related to complications. They implicated all major aortic surgery (including endovascular techniques) with development of a fistula any time after primary surgery. The longest delay they discovered was 26 years. They concluded that bleeding was the dominating symptom and that diagnostic delay, although the norm, has decreased over time. Overall mortality, although high, has also decreased over time. It is further decreased following revascularisation and aortic graft removal.¹

Thus the rarity of this condition as well as its often non-specific presentation and high mortality make it an important one for discussion and consequent dissemination to medical practitioners of various specialties. This case highlights the role of careful history and clinical examination. In addition, it reinforces the limitations of CT angiography which requires active bleeding for a positive result and can therefore lead to a false reassurance when negative.

CASE PRESENTATION

A female in her sixties presented with haematemesis, malaena and a significant drop in haemoglobin on three separate occasions over a two-month period. Her initial medical history was that of only hypertension. On her third admission a catastrophic bleed resulted in emergency surgery and ultimately her death.

It presented in an unusually long period in this particular case. The patient had undergone an aortic endarterectomy with primary suture closure

of the aortic wall at a neighbouring hospital some six years prior but this aspect of her history was unfortunately overlooked until her final admission.

INVESTIGATIONS

Numerous upper gastrointestinal endoscopies were carried out during her three admissions. Many of these revealed fresh blood in the stomach but no cause was identified despite intubation of the third part of the duodenum. A colonoscopy was attempted but abandoned in the sigmoid because of poor views from extensive malaena. Her initial CT angiogram did not reveal any abnormality. Her second CT angiogram was carried out during a significant bleed and demonstrated active extravasation of contrast from the anterior aorta in to the third part of the duodenum (figure 1). The aorta was normal in calibre and there was no graft material visible on the CT in keeping with a previous suture repair of the aorta.

DIFFERENTIAL DIAGNOSIS

Haemobilia: This is the consequence of bleeding into the biliary tree. It is another uncommon cause of obscure upper gastrointestinal (GI) bleeding. The usual mechanisms for this phenomenon are trauma, pseudoaneurysm, atherosclerotic aneurysm or vascular malformations although cases secondary to chronic cholangitis have also been described.²

Haemosuccus pancreaticus: This is the rare phenomenon of upper GI bleeding via the ductus pancreaticus major or minor (satorinirrhagia). The mechanisms for this are: rupture of an aneurysm/pseudoaneurysm of a peripancreatic artery into the pancreatic duct or into a pancreatic pseudocyst communicating with the duct, trauma, iatrogenic (eg, during endoscopic retrograde cholangiopancreatography), pancreatitis, pancreatic cancer, pancreaticolithiasis, vascular malformations or pancreas divisum.³

DISCUSSION

Aorto-enteric fistulas are classified as either primary or secondary. Primary fistulas, arising in an unoperated or so-called 'virgin' aorta are rare.⁴ Secondary fistulas occur after previous aortic surgery most commonly where a prosthetic graft has been inserted but can also occur at suture lines in the absence of graft material as in our case.

The underlying aetiology is thought to relate to chronic low-grade infection around the graft and subsequent erosion, most commonly into the third part of the duodenum.⁵ Upper GI endoscopy can

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Figure 1 (A) Transverse and (B) sagittal CT angiogram images demonstrating extravasation of contrast from the aorta into the third part of the duodenum.

be used to investigate but only reveals the fistula in a minority of cases.⁶

Typically the patient presents with a herald bleed at which point the patient is relatively stable and clot formation at the fistula site prevents further bleeding. This is usually followed by a catastrophic bleed most often within hours to days. Our case was unusual in that our patient suffered numerous herald bleeds over 2 months before presenting with a huge bleed. Her initial CT angiogram had been negative but was performed during a period when she was not actively bleeding. For CT angiography to demonstrate a bleeding site with contrast extravasation there needs to be active bleeding at a rate of approximately 0.5 ml/min.⁷

Open repair has long been regarded as the first line in treatment. The two main concerns intraoperatively relate to adequate control of bleeding and prevention of recurrence or infection in the future. There are a number of operations available all of which commence with resection of the abnormal aorta. Subsequent reconstruction may involve axillo-femoral bypass or in situ aortic grafting with a standard prosthesis,

autogenous femoral vein graft and even a cryopreserved autograft. Repair (<3 cm) or resection (>3 cm) of the gastrointestinal lesion should then be undertaken together with omental or peritoneal flap interposition recommended to prevent recurrence.⁸

Recently endovascular aorto-enteric fistula repair (E-AEFR) has been postulated as an alternative to open repair.

Research has demonstrated that in-hospital mortality and morbidity are lower in E-AEFR than open repair. However, the early benefits of E-AEFR may be outweighed by an increased risk of long-term recurrence and infection compared to open surgery.⁹

Therefore it can be seen that while there is a role for endovascular repair, further studies are required to assess its long-term outcomes. Currently it seems that it may be used as a bridging procedure allowing definitive surgical repair under more controlled circumstances.^{10 11}

Learning points

- ▶ Aorto-enteric fistulas are a rare cause of gastrointestinal (GI) bleeding and most commonly follow aortic surgery.
- ▶ Meticulous history and clinical examination are critical steps in making a diagnosis.
- ▶ Normal CT angiography studies need to be interpreted with an understanding of their limitations.
- ▶ A herald bleed typically precedes the catastrophic bleed by a period of hours to days but occasionally longer.
- ▶ Upper GI endoscopy can be used to demonstrate a fistula in the third part of the duodenum but will often not visualise it despite careful examination.

Competing interests None.

Patient consent Obtained.

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