Case Report of Secondary Aortoenteric Fistula: A Rare Cause of Upper Gastrointestinal Bleeding

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Abstract

Introduction: Upper gastrointestinal bleeding is common and may be mild to potentially life threatening. The commonest cause is ulceration and variceal bleeding, however there are many other less common causes. Endoscopy is the mainstay of diagnosis and also often allows for concurrent management. In some cases though, endoscopy alone may not be sufficient to identify the underlying etiology.

Case Presentation: The case describes a 70-year-old male who developed upper gastrointestinal bleeding 13 days after cholecystectomy complicated by bile leak requiring ERCP and stenting. His background was significant for open AAA repair complicated by chronic graft infection. Initial management was based on a presumed diagnosis of stress ulceration or post-sphincterotomy bleed with endoscopy showing friability of the periampullary mucosa. However based on CT findings and clinical history the patient was diagnosed with secondary aortoenteric fistula. He underwent surgical bypass and repair.

Discussion: Aortoenteric fistula is a rare cause of upper gastrointestinal bleeding. It may present with a herald bleed or with massive haemorrhage. Diagnosis may be difficult with the diagnosis often not able to be made by endoscopy. High index of suspicion based on patient history is critical, as well as CT findings. Initial management is resuscitative followed by extranatomical bypass, resection of the affected graft and repair of the affected small bowel.

Conclusion: Upper gastrointestinal bleeding is common, while aortoenteric fistula is an uncommon but potentially life-threatening cause. Early diagnosis and management is crucial and requires a high index of suspicion as well CT findings and endoscopy.

Keywords: Abdominal aortic aneurysm; Aortoenteric fistula; Case report; CT findings; Upper gastrointestinal bleeding

Abbreviations: AAA: Abdominal Aortic Aneurysm; ERCP: Endoscopic Retrograde Cholangiopancreatography; CT: Computed Tomography

Introduction

Upper gastrointestinal bleeding is common, with incidence of 61 people per 100 000 in 2009 globally. It may vary in severity from mild to potentially life-threatening with mortality reaching up to 11% [1]. The most common causes include peptic ulcer disease related to H. pylori and non-steroidal anti-inflammatories, and variceal bleeds in cirrhotic patients [2]. Less common causes include Dieulafoy lesions, haemobilia, gastric antral vascular ectasia, arteriovenous malformations and aortoenteric fistula [3]. The mainstay of both diagnosis and management is upper endoscopy however in a small amount of cases this alone may not be adequate to determine the cause and effectively treat the problem. The following case from a regional secondary referral hospital describes a patient with a rare cause of upper gastrointestinal bleeding and emphasises the need for attention to clinical history and important CT findings to allow for identification and optimal definitive management.

Case Presentation

A 70-year-old man underwent delayed cholecystectomy following cholecystostomy for calculous cholecystitis while on dual antiplatelet therapy following NSTEMI. His background was significant for coronary artery disease, open AAA repair 8 years prior complicated by chronic graft infection on long term antibiotic therapy, and cavitating lung disease. Intra-operatively he was noted to have omental adhesions to the gallbladder, as well as a mucous fistula from the fundus to the anterior abdominal wall.
Despite this, Calot’s was dissected and the critical view of safety obtained. Intra-operative cholangiogram performed which was noted to be normal. The patient developed severe abdominal pain with guarding day one post-operatively and returned to theatre for diagnostic laparoscopy. He was found to have bile peritonitis with possible bile leakage from gallbladder bed. A decision was made to washout and place a drain in the gallbladder fossa with view to perform subsequent ERCP. ERCP demonstrated a bile leak from the middle third of the common bile duct. Sphincterotomy was performed and a plastic stent placed. The patient was then transferred to the nearest centre with hepatobiliary surgery. During his stay, no further treatment was required, and the patient went home thirteen days after his original operation.

The following day the patient represented to the emergency department severe abdominal pain and haematemesis. He was noted to be hypertensive with guarding in the epigastrium and right upper quadrant. The patient was transfused packed red cells, started on octreotide and pantoprazole, and given tranexamic acid. He underwent CT which was reported to show a distended duodenum with bleeding which appeared to be originating from the duodenal wall. There was also noted to be an incidental finding of gas in the aorta of which the significance was unknown in setting of chronically infected aortic graft. Upon discussion between surgery and gastroenterology it was thought the source of bleeding was most likely either a stress ulcer or post-sphincterotomy bleed and the patient was taken for endoscopy. Clotted blood was noted in both the stomach and the duodenum. Localised mucosal changes characterised by congestion, friability and ulceration were found at the ampulla, presumable from the recent ERCP and sphincterotomy. Coagulation using heater probe and injection of adrenaline were both unsuccessful, however haemospray appeared to result in successful haemostasis.

The patient was transferred to intensive care and had a subsequent 6 units of packed cells transfused over the following day. He also proceeded to have two further episodes of haematemesis. The patient’s bilirubin increased to 200/138 and there was concern for both ongoing bleeding and potentially a blocked stent. A decision was made to take the patient back for repeated endoscopy. The stomach and duodenum were again noted to be full of clotted blood. The likely bleeding point was identified at the ampulla with blood noted to be trickling down the bile duct. The previously placed plastic stent was noted to have migrated out of the duct into the duodenum and was replaced with a covered metal stent with apparent control of the bleeding. The case was further discussed with the hepatobiliary surgeon at the closest tertiary centre. It was thought possible that the bleeding may have been due to stent irritation of the periampullary mucosa, however this was unlikely to cause significant transfusion and the patient had now developed a new inotropic requirement with ongoing bleeding. On further review of the imaging further concern was raised regarding the locule of gas in the aorta, raising the possibility of aortoenteric fistula. The case was discussed with the vascular surgery team and a decision was made to perform combined vascular and general surgery procedure.

The patient underwent extra-anatomical axillofemoral bypass using 8mm reinforcedortex graft. Laparotomy identified the proximal aortic graft plastered to the third part of the duodenum with three separate holes noted in the duodenum. The duodenum was mobilised off the graft and the aorta tied off proximally and distally. The infected graft was then removed with extensive washout of the retroperitoneum. Given the physiological state of the patient and the fact there was three separate holes in the duodenum it was decided that it would not be possible to perform three separate primary repairs. Therefore, decision was made to perform a stapled side to side anastomosis well away from D2. A feeding jejunostomy was placed distal to the anastomosis. The patient discharged 18 days post-operatively and noted to be recovering well at the time of clinical review three months later. He remains well in the community 2 years after this event.

**Discussion**

Aortoenteric fistula is a rare entity which can occur either as a primary issue or secondary to previous abdominal aortic aneurysm repair. Patients typically present with upper gastrointestinal bleeding which may be minor, typically known as a herald bleed, or potentially life threatening [4]. Given the scarcity of these lesions a high index of suspicion in any patient with known aortic aneurysm or previous abdominal aortic aneurysm repair is required to correctly diagnose aortoenteric fistulas to allow for early management. CT angiogram can also be used to aid diagnosis with features of aortoenteric fistula including ectopic gas adjacent to or within the aorta, focal bowel wall thickening, discontinuity of the aortic wall and extravasation of contrast into the bowel lumen [5]. Upper endoscopy may assist in diagnosis however sensitivity of endoscopy for diagnosing aortoenteric fistula is only about 50% [6]. Mortality from aortoenteric fistula ranges from 14 to 70% with most patients dying before the diagnosis of aortoenteric fistula is made [7]. Initial management is resuscitative involving transfusion and haemostasis with either cross clamping the aorta or endovascularly with REBOA. Definitive management requires explantation of the graft, extra-anatomical bypass of blood supply and repair of the affected region of small bowel.

**Conclusion**

Upper gastrointestinal bleeding is a common presentation with many possible differentials. Determining the correct source may sometimes be difficult but management may vary significantly depending on the cause. Close attention should be given to the patient’s clinical presentation and past medical history with a high index of suspicion. Useful adjunct investigations include
both upper endoscopy and CT imaging. Early diagnosis and management offer the best chance of a good outcome in patients with aortoenteric fistula which has a high associated mortality.

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