CASE REPORT

A rare and forgotten diagnosis of gastrointestinal bleeding: primary aortoduodenal fistula

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SUMMARY

Upper gastrointestinal (UGI) bleeding is a common medical condition among adults. Bleeding is mainly caused by the gastric or proximal duodenum, but rarely by the distal duodenum. Aortoduodenal fistula is one of the causes of UGI bleeding that results in a life-threatening condition if not treated properly. It is commonly observed in patients who have undergone previous aortic surgery and rarely occurs in patients without a history of aortic surgery. Diagnosis of aortoenteric fistula is difficult unless there is a high level of clinical suspicion. In this article, we aim to present a patient with aortoduodenal fistula and discuss the clinical presentation, as well as the diagnostic and therapeutic options of this disease.

BACKGROUND

Upper gastrointestinal (UGI) bleeding is a common medical condition that results in a high level of patient morbidity-mortality and medical care costs. In patients with a clinical presentation of haematemesis/melaena, diagnostic work is often performed after haemodynamic stability and resuscitation are achieved. The use of UGI endoscopy greatly facilitates the diagnosis and initial treatment of frequently encountered aetiologies such as peptic ulcers, oesophagogastric varices and tumours.¹ However, diagnosis with endoscopy is difficult for pathologies that are located in the distal part of the duodenum.

Among these pathologies, aortoduodenal fistula (ADF) is a rare cause of UGI bleeding that results in a life-threatening condition with 100% mortality if not treated properly.² This greatly increases the importance of the treatment algorithm. Although it is commonly observed in patients with previous aortic reconstruction surgery (secondary ADF), it can also occur in patients with no history of aortic surgery (primary ADF). This disease, for which a rapid treatment approach following clinical suspicion is key to saving the patient, is commonly observed in cases with grafts; hence, clinical suspicion for primary ADF generally remains in second plan.³ However, it is important to bear this condition in mind for patients with no distinct findings in UGI endoscopy that elucidates the cause of bleeding. In this article, our aim is to describe the clinical presentation, diagnosis and treatment approach for a patient admitted for melaena and later identified with primary ADF following various examinations and tests.

CASE PRESENTATION

A 55-year-old man was admitted to our department with epigastric pain and melaena. Abdominal examination revealed no symptoms other than mild epigastric tenderness. His initial vital signs revealed blood pressures in the normal range and his heart rate was tachycardic. Important laboratory values for the patient were identified as follows: haemoglobin level, 9.6 g/dl; haematocrit, 29.2%; platelet count, 280 000/ml and the international normalised ratio, 1.49. After fluid resuscitation and haemodynamic stability were achieved, the patient was hospitalised with a diagnosis of UGI bleeding. Patient history revealed that the patient had been admitted to a hospital 10 days previously with clinical manifestations of shock and for UGI bleeding. Following tests and blood replacement, the patient had been discharged after receiving treatment for peptic ulcer. The patient also had a history of myocardial infarction that had occurred 3 years previously, for which he was receiving antiaggregant treatment. Patient history revealed no other diseases or previous surgeries. No pathology was identified in the patient's UGI endoscopy. No sites of bleeding or clots were observed inside the stomach or the duodenum.

INVESTIGATIONS

Following a decrease in the haemoglobin values of the haemodynamically stable patient, and a continuation of melaena, CT angiography was performed. The CT identified no active bleeding in the patient; however, an aneurysm was observed with a diameter of 62 mm in the abdominal aorta, containing a mural thrombosis of nearly 12 mm. Also, effacement of the fat plane between the aorta and the adjacent portion of the duodenum and intra-duodenal contrast material was observed (figure 1). Based on these findings, surgery was performed with a preliminary diagnosis of ADF.

TREATMENT

Formations of an aneurysmatic dilation and an inflammatory mass between the aorta and the 3rd–4th portions of the duodenum were observed intraoperatively. After clamping the proximal and distal sections of the aorta, the duodenum was detached from the aorta, and a 6–8 mm fistula tract was identified in the region. Following a wedge resection and primary duodenography performed on the duodenum, the aorta was reconstructed with an aortobiliac graft (figure 2).

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Figure 1 Axial (A), coronal (B), sagittal (C) and three-dimensional volume rendered (D) images show abdominal aortic aneurysm containing a mural thrombosis, effacement of the periaortic fat plane, bowel wall thickening and a small amount of contrast medium in the duodenum (arrow).

Figure 2 An opened sac revealing aortoduodenal fistula in the third part of the duodenum (A) and a view of the operative field after reconstruction (B).

OUTCOME AND FOLLOW-UP

No bacterial growth was observed in the cultures that were obtained preoperatively. After an uneventful postoperative period, the patient was discharged on the 14th day following surgery.

DISCUSSION

Aortoenteric fistula is a rare cause of gastrointestinal bleeding that is associated with high mortality. Although it is most commonly observed between the aorta and the duodenum, fistulas may also develop between the aorta and the oesophagus, the jejunum and the sigmoid colon. ADFs are divided into two groups depending on the presence or absence of previous aortic surgery. Aortoenteric fistulas are more commonly observed as a late onset complication of aortic surgery. On the other hand, primary ADF, first described by Sir Astley Cooper, is a rare occurrence of which less than 500 cases have been reported in the literature.⁴ Whether or not the reported number of cases reflects the actual incidence of primary ADF is arguable. Real incidence is likely to be much higher either due to the inability to diagnose the condition in most patients, or due to the cases that are not reported.⁵

Although primary ADF is frequently observed as a complication of an aneurysm, it may also develop as a result of infection, tumour, trauma, radiotherapy or foreign objects. In case an underlying cause of primary ADF is present, it will lead to the erosion of the aorta and to its connection with adjacent organs. The subsequent contamination and infection by the intestinal flora will cause persistence of this connection.⁶ Such fistulas generally develop in the region between the aorta, the superior mesenteric artery and the third portion of the duodenum. In this report, we discuss a case of ADF that has developed following an aneurysm of the aorta.

Although gastrointestinal bleeding, abdominal pain and a pulsating abdominal mass are described as the classical triad of ADF, it is only in 11% of patients that all three of these symptoms are observed at the same time. Gastrointestinal bleeding is frequently observed as a haematemesis, a melaena and even a haematochezia. In addition, back pain, episodes of syncope and shock are other symptoms that may be observed. The classical triad was not observed in the case that was discussed, and the patient's clinical presentation also included melaena and episodes of syncope. Another finding as significant as these observations is the presence of 'herald bleeding'. Observed in nearly 30% of patients, it is a form of bleeding that usually precedes a massive haemorrhage. After causing hypotension, vasospasm or the obstruction of the fistula with a thrombus in the patient, an episode of herald bleeding ends as spontaneously as it begins. The interval between a herald bleeding and the subsequent haemorrhage may vary from several hours to several months, but in 50% of patients, this interval is more than 24 h.⁵ This presented patient had been admitted to the hospital 10 days previously with the symptom of herald bleeding; however, he was discharged after clinical improvement.

UGI endoscopy is the first diagnostic approach for patients with a clinical presentation of UGI bleeding. However, as the bleeding in ADF is generally located in the distal duodenum, endoscopy's sensitivity to ADF is relatively low. Nevertheless, a UGI endoscopy should still be employed as the first diagnostic method in order to exclude other causative factors. It is important to bear in mind that negative findings are as important as positive findings, and that diagnosis may require additional tools.⁷ While angiography, scintigraphy and barium-contrasted

methods are insufficient for the diagnosis of ADF in the absence of massive bleeding, abdominal CT is the most valuable diagnostic tool, with a sensitivity of 50–94% and a specificity of 85–100%.² For CT evaluation, the most specific finding suggests that ADF is the direct extravasation of contrast material from the aorta into the intestinal lumen, or the extravasation of enteric contrast material into the periaortic region. However, these findings are not frequently encountered, and the diagnosis of ADF is generally performed based on an evaluation of the indirect findings. The effacement of the periaortic fat planes, the presence of periaortic fluid or air, and the thickening of the bowel loops adjacent to the aorta are the commonly observed findings. Based on an evaluation of these findings, a differential diagnosis from retroperitoneal fibrosis, infected aorta aneurysm or infectious aortitis should be evaluated.⁸ In the presented case, the extravasation of contrast material from the aorta to the duodenum, the effacement of the fat planes between the frontal wall of the aorta and the duodenum, and the thickening of the intestinal walls were findings that were suggestive of ADF.

Considering that a wait and see approach would result in the patient's death, surgery is a definite approach for a patient assumed to have ADF, both for diagnosis and treatment. During exploration, following the control of the proximal and distal parts of the aorta, the fistula should be disconnected. Primary repair of the aorta and the duodenum, resection of the aneurysm and reconstruction with grafts, in situ grafts and an extra-anatomic bypass are several methods of different reconstructive techniques that may be used.⁹ Primary repair or resection and anastomosis of the duodenum are all methods that can be employed; however, the technique that is used for the reconstruction of the aorta is more important. One of the most important causes of postoperative mortality is sepsis which develops due to anastomosis leakage and graft infection. For this reason, pre-operative or postoperative microbial culture studies and the assessment of periaortic infection are quite important. The presence of severe infection is associated with high morbidity and mortality; however, grafts may be used in the absence of pus. In the presented case, aortic reconstruction with a graft was performed because of the absence of local infection and no complication was observed in the postoperative period. In clinically unstable patients, bridge procedures with angiographic embolisation or endovascular plugs is an alternative approach that may be performed prior to definitive treatment.¹⁰

In conclusion, despite ADF being a rare cause of gastrointestinal bleeding, the fact that it results in high mortality when left untreated means that physicians in the emergency department assume a major role in the prognosis of patients. Regardless of whether patients do or do not have a history of aortic surgery,

Learning points

- ▶ Aortoduodenal fistula (ADF) is a rare cause of gastrointestinal bleeding. It must be included and be remembered within the differential diagnoses of all cases of upper gastrointestinal (UGI) bleeding.
- ▶ Mortality owing to ADF is very high when left untreated.
- ▶ Regardless of whether patients do or do not have a history of aortic surgery, the possibility of ADF should be considered for patients with UGI bleeding.
- ▶ Endoscopy has very low sensitivity to reveal aortoduodenal fistula.

the possibility of ADF should be considered, and must be remembered within the differential diagnoses of all cases of UGI bleeding, especially for those with negative endoscopic findings. Also, the essential diagnostic and therapeutic approaches should be followed accordingly.

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