CASE REPORT

Haemobilia due to hepatic artery pseudoaneurysm

Cristina Teixeira, Suzane Moura Ribeiro, Ana Luisa Alves, Isabelle Cremers

Gastroenterology, Centro
Hospitalar de Setúbal EPE,
Setúbal, Portugal

Correspondence to

Dr Cristina Teixeira,
ac.corda.teixeira@gmail.com

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SUMMARY

An 83-year-old woman with a history of hepaticoduodenostomy 20 years ago was admitted with upper gastrointestinal bleeding. Emergency upper gastrointestinal endoscopy revealed multiple blood clots over the stomach and first and second parts of the duodenum. The cannulation of the biliary tree with a flexible end-viewing endoscope exposed the presence of blood clot inside biliary lumen and a semipedunculated polyp which, at first, appeared to be the cause of haemorrhage. A few days after polypectomy, patient was discharged home, however, was admitted again with massive bleeding and selective angiography demonstrated a pseudoaneurysm of left hepatic artery. Angioembolisation was performed and haemorrhage was stopped afterwards.

BACKGROUND

Haemobilia is an uncommon condition that occurs when a fistula is formed between a vessel of the splanchnic circulation (hepatic artery or portal vein) and the intrahepatic or extrahepatic biliary system.¹

The classical triad of symptoms described by Heinrich Quincke in 1871,² which includes pain, jaundice and gastrointestinal bleeding, is pathognomonic of haemobilia. Nonetheless, the triad is present in only 22% of the patients.¹

While in the past the major causes for haemobilia were blunt or penetrating liver trauma, nowadays, in the majority of cases, the most common cause is iatrogenic liver trauma, secondary to surgical, endoscopic or radiological procedures.³ In these circumstances, haemobilia can be the result of a traumatic arteriobiliary fistula or a pseudoaneurysm, which accounts for 10% of haemobilia cases,⁴ and may present with delayed bleeding.⁵ Other causes of haemobilia include inflammatory disorders such as hepatic abscesses, pancreatic pseudocyst, cholangitis and cholecystitis, coagulopathy, real aneurysms and gallstone disease, as well as primary and secondary malignancies of the liver, bile duct, gallbladder and pancreas.^{1 6 7} The interval between the triggering event and onset of haemobilia is variable and may be several weeks to months.¹

Diagnostic options for haemobilia include endoscopy, angio-CT scan, ultrasonography or angiography. Besides making the diagnosis, angiography has the capability of therapeutic intervention; embolisation has a high success rate and represents the first line procedure to stop bleeding.^{6 8} Classic surgical interventions should be considered only when embolisation fails, since they carry high morbidity and mortality rates.⁹

The management of haemobilia is aimed at stopping the bleeding, maintaining continuous flow through the biliary system and treating the underlying aetiology.¹⁰

We report the clinical history of a patient who presented with massive recurrent upper gastrointestinal haemorrhage due to a pseudoaneurysm of left hepatic artery and discuss the confounding issues that were encountered in the diagnosis and management.

CASE PRESENTATION

An 83-year-old woman was admitted at the emergency department after an episode of sharp abdominal pain and haematochezia. Her medical history included cholecystectomy 30 years ago and, 20 years ago, due to residual lithiasis, a biliodigestive derivation surgery (hepaticoduodenostomy).

On physical examination at admission she was pale; blood pressure was 142/82 mm Hg, pulse rate was 102/min and respiratory rate was 22/min. Epigastric and right upper quadrant tenderness was noted with no palpable abdominal mass or organomegaly, and digital rectal examination revealed melena. Blood clots were also aspirated following the insertion of a nasogastric tube. Blood tests revealed anaemia (haemoglobin 9.3 g/dL), leucocytosis (12×10^3 cells/mm³) with neutrophilia (80%), elevated C reactive protein (CRP) of 7.95 mg/dL and abnormal liver tests with alanine aminotransferase of 183 U/L, aspartate aminotransferase of 296 U/L, alkaline phosphatase of 487 U/L, gamma-glutamyl transpeptidase of 309 U/L, total bilirubin of 1.79 mg/L with a direct fraction of 1.2 mg/dL and prothrombin time of 15.3 s. Renal profile and electrolytes were normal.

In the following hours, haemodynamic instability developed followed by the passage of red bloody stools and a decreasing haemoglobin level to 7.2 g/dL. Although the patient did not present jaundice, repeated laboratory tests also revealed increasing bilirubin level, which reached up to 3.1 mg/dL. The patient was stabilised with fluid replacement and transfusion of blood units and underwent oesophagogastroduodenoscopy (OGD) that demonstrated fresh blood and clots in the stomach and duodenum and a biliodigestive anastomosis in the first portion of the duodenum. A cholangioscopy through the anastomosis, to the common hepatic duct, revealed a blood-filled biliary tree and after removal of clots, a pedunculated polyp (figure 1) was visualised inside the biliary duct, with slow bleeding, which stopped after injection of adrenalin and polypectomy was performed. Histopathological examination revealed duodenal mucosa with hyperplastic epithelium and oedema.

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Figure 1 Polyp found in the common hepatic duct.

Following the procedure, there was no additional bleeding, the patient remained stable and 3 days later was discharged. One week later, the patient was readmitted, complaining of fatigue and melena. Her haemoglobin level had dropped from 10 g/dL to 5.9 g/dL. A new OGD was carried out but failed to identify any active bleeding lesion, showing only a small ulcer from polypectomy (figure 2). The patient remained asymptomatic after blood transfusion but 3 days later, she experienced an episode of haematemesis. A third OGD clearly showed active bleeding from the biliary tree. A selective angiography of celiac trunk was arranged in another institution and disclosed a 2 cm pseudoaneurysm in dependency of a segmental branch of left hepatic artery (figure 3), in segment II. She underwent selective microcoils (2–4 mm) embolisation to occlude the feeding artery both proximal and distal to the pseudoaneurysm with successful aneurysmal exclusion (figure 4). Bleeding ceased thereafter and no additional blood transfusion was required. However, a few hours after intervention, she presented with high fever (38.3°C), epigastrium pain and a peak in white blood cell count and CRP. CT scan of the abdomen revealed a low density area with 33 mm in diameter in segment II of the liver, suggesting a hepatic infarction of this segment. Antibiotics and support treatment were initiated, oral feeding was allowed on the third day

Figure 2 Small ulcer postpolypectomy in the biliary ducts.

Figure 3 Pseudoaneurysm demonstrated by angiography.

of intervention and patient was discharged 1 week later with no impairment of liver function tests.

OUTCOME AND FOLLOW-UP

Recovery was uneventful with no signs of haemobilia recurrence and with normal laboratory profile after a 6-month period of follow-up.

DISCUSSION

Massive haemobilia is a rare but potentially life-threatening condition. This entity may be difficult to recognise but it should be included in the differential diagnoses of upper gastrointestinal bleeding (UGB).

Acute biliary symptoms and signs combined with acute UGB should strongly alert the clinician to the possibility of this diagnosis.¹¹ In our case, two of three symptoms of the Quincke's triad (bleeding and abdominal pain) were initially present and even though clinical jaundice was not recognised, there were laboratory changes suggesting acute biliary obstruction.

Haemorrhage into the biliary tract occurs when trauma or disease produces a fistula between a vessel of the splanchnic

Figure 4 Selective microcoils embolisation with aneurysmal exclusion.

circulation and the bile ducts, either in the intrahepatic or the extrahepatic biliary tract.¹² Our patient had been submitted in the past to two surgical interventions that were probably related with the development of this pseudoaneurysm complicated by massive haemorrhage. Postcholecystectomy haemobilia usually occurs as a result of cystic or hepatic arterial wall injury, which can induce a traumatic arteriohepatic fistula or the development of a pseudoaneurysmatic lesion that may manifest itself with delayed bleeding into the biliary tree.³ The exact mechanisms responsible are not well understood; however, direct vascular injury during surgery, electric current diffusion through clips placed close to the vascular pedicle and vascular erosion due to clip encroachment may have a role in the creation of hepatic artery pseudoaneurysm.¹³

The diagnosis of haemobilia is not easy to establish and often a long period elapses between the iatrogenic procedure to the onset of the symptoms and finally to the identification of a source of bleeding.¹² Brisk bleeding into the duodenum or a slow blood flow leading to formation of blood clots may occur, obstructing the bile ducts and causing jaundice, abdominal pain or abnormal liver tests.⁶ Acute biliary obstruction by clot may also cause cholangitis, cholecystitis or pancreatitis.¹⁴ Occasionally, as with our patient, the haemorrhage may be recurrent.¹

An endoscopy is diagnostic of haemobilia in only 10% of cases, which increases if the patient presents UGB.¹⁵ In our patient, an altered postoperative gastrointestinal anatomy and the presence of active bleeding in an identified lesion impeded earlier establishment of the diagnosis and thus misdirected the therapeutic strategy in the beginning. Contrast enhanced CT may be used after UGB as an initial study to obtain additional information when haemobilia is suspected; however the site of bleeding cannot be accurately located by this modality.¹ Although this examination will not show the erosive vascular changes as seen on arteriograms, it may unveil haemorrhage or pseudoaneurysm formation.¹³ Interventional angiography is regarded as the 'gold standard' for diagnosing haemobilia and treatment of the underlying cause of bleeding.⁵ Transarterial embolisation (TAE) is now the first line of intervention to stop bleeding, as its reported success rate ranges from 80% to 100%.⁶

Initial management of a patient with clinical suspicion for haemobilia must begin with appropriate goal-directed resuscitation and correction of coagulopathy. If biliary obstruction is present, appropriate drainage must be ensured after bleeding is addressed. Percutaneous transarterial angiography by an experienced hepatopancreatobiliary interventional radiologist is critical both for the diagnosis and treatment of haemobilia. The embolisation of vascular lesions with materials such as microcoils, gelatin sponge, polyvinyl alcohol or cyanoacrylate achieves haemostasis in 75%–100% of patients.¹⁶ It has advantages over surgical approaches: it enables treatment in a single session and can also be repeated, it is safer because it deals directly with the arterial lesion and it is better tolerated in patients who are haemodynamically unstable and therefore at high anaesthetic and surgical risk.⁶

Our patient presented hepatic infarction after TAE. In fact, the procedure, although safer than surgery, is not without risk. Hepatobiliary necrosis (6%), abscess formation (9%), bleeding (6%) and gallbladder fibrosis (2%) have occurred following arterial embolisation¹⁴ and sometimes are related to incorrect

selection of embolic agents and inappropriate use of superselective technique.¹⁷ Surgery is indicated when TAE has failed, in cases of hepatic sepsis or haemobilia associated with cholelithiasis, cholecystitis or resectable neoplasms.¹⁴ Surgery for haemorrhage involves ligation of the bleeding vessel or excision of the aneurysm.¹⁸

Learning points

- ▶ Haemobilia is rare and the diagnosis on a high index of awareness, especially when the common causes of upper gastrointestinal tract haemorrhage have been excluded.
- ▶ A clinical suspicion should prompt confirmation with angiography and endoscopy, but CT scan or ultrasonography may also be helpful.
- ▶ Transarterial embolisation is the first line of intervention to treat haemobilia.

Contributors CT, SMR and IC followed the patient. CT and SMR wrote the article. IC and ALA revised the article.

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