CASE REPORT PEER REVIEWED | OPEN ACCESS

Hemobilia after laparoscopic gallbladder bed resection and portal lymphadenectomy for incidental gallbladder adenocarcinoma

José Pedro Gonçalves, Pedro Maurício Valente, Pedro Filipe Sousa, Francisco Cocco, Gil Faria

ABSTRACT

Introduction: Upper gastrointestinal bleeding caused by hemobilia is rare and a consequence of vascular-biliary fistula. These fistulas are mainly associated with biliary surgeries' iatrogenic injuries.

Case Report: This case reports of an elderly man with prior history of laparoscopic gallbladder bed resection and portal lymphadenectomy, which presented with hemobilia one month after the surgery. Diagnosis was not straightforward, but the patient was successfully treated with selective arteriography and hepatic artery stenting.

Conclusion: High suspicion for hemobilia in a patient with right upper quadrant pain, jaundice, and blood loss in the stools after biliary surgery and rapid intervention are essential to avoid further complications.

Keywords: Anemia, Arteriography, Cholecystectomy, Gastrointestinal bleeding, Hemobilia

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INTRODUCTION

Gastrointestinal (GI) bleeding is a frequent cause of hospitalization. Usually defined as upper GI bleeding when it occurs above the ligament of Treitz and as lower GI bleeding if it happens distal to that ligament. Hemobilia is one of the rarest and seldom forgotten causes of upper GI bleeding.

First described in 1654 by Glisson, and named by Sandblom in 1948 [1], hemobilia is defined as: blood flow through the biliary tree leaving the duodenal papilla and manifesting as melena (or hematochezia when higher flow is present). It is the consequence of an arterio-biliary fistula. The high arterial pressure forces blood flow into the low-pressure biliary ducts, on the other hand, if there is a venous-biliary fistula, bile may enter and mixture with blood leading to bilhemia, with a rapid and high raise in bilirubinemia [2, 3].

Most commonly, a vascular-biliary fistula is an iatrogenic lesion which is caused either by laparoscopic or classic cholecystectomy, percutaneous

transhepatic cholangiography, endoscopic retrograde cholangiopancreatography (ERCP), liver biopsy, other liver surgeries or blunt trauma to the liver. Less common causes are cholelithiasis, biliary tumors and chemoradiation therapy to intrahepatic tumors, infections, vascular diseases, and aneurisms [4–8].

The classic symptoms of hemobilia are Quincke's triad: jaundice, right upper quadrant colicky pain, and GI bleeding. This triad is found in about 22–40% of the patients [4, 9, 10]. When blood clots obstruct the bile ducts, jaundice and pain worsens. Otherwise, pain usually alleviates after blood loss [11].

It is important to exclude hemobilia in patients presenting with GI bleeding after biliary surgery. Misdiagnosing it can lead to severe hypovolemic shock and death.

CASE REPORT

A 76-year-old man was admitted to the general surgery emergency room (ER) with epigastric pain, nausea, jaundice, and melena for the previous three days.

He had a past medical history of diabetes mellitus controlled with oral antidiabetic agents, appendectomy and treated prostate cancer. He had been submitted to laparoscopic cholecystectomy for a polyp, which on histological examination revealed a gallbladder adenocarcinoma (pT2N0Mx). He was evaluated at the multidisciplinary tumor board and proposed for completion surgery. He underwent laparoscopic gallbladder bed hepatic resection and portal lymphadenectomy 37 days prior to ER presentation. During this surgery, there was an iatrogenic puncture of the main bile duct, which was sutured with 4-0 polypropilene suture. The post-operative was uneventful.

On physical examination the vital parameters were normal. His abdomen was soft and non-tender, with epigastric and right hypochondrium pain at deep palpation, without peritoneal signs. Digital rectal examination was positive for melena.

Blood tests at admission revealed: Hemoglobin was 12.9 g/dL; white blood cell (WBC) count, platelets, and bleeding times were normal; total bilirubin was 4.8 mg/dL; conjugated bilirubin was 2.9 mg/dL; aspartate transaminase (AST) was 342 U/L; alanine transaminase (ALT) was 777 U/L; gamma-glutamyltransferase (γ-GT) was 1485 U/L; alkaline phosphatase (ALP) was 655 U/L; C-reactive protein (CRP) was 23.7 mg/dL.

The computed tomography (CT) scan revealed dilatation of the extra-hepatic bile duct with a partial obstruction of the distal segment, hypothesizing biliary stones, or a clot inside the duct. He was diagnosed with cholangitis and admitted to treatment with intravenous antibiotics and endoscopic retrograde cholangiopancreatography. After ERCP with sphincterotomy and removal of blood clots the patient had hematochezia and a drop in hemoglobin to 8.7 g/dL. An upper endoscopy was performed and showed an adherent clot to Vater's papilla, next to the sphincterotomy, non-washable and without active bleeding. Four days later a new episode of hematochezia occurred. The patient presented hemodynamic instability, with a significant fall in hemoglobin to 5.9 g/dL, thus requiring transfusion of 2 units of packed red blood cells. Since bilirubin and cholestasis kept increasing, a new ERCP was done to stent the bile duct, during which hemobilia was observed.

A trial of conservative treatment with aminocaproic acid and octreotide was successful and the patient was discharged seven days later, without any further blood losses and a raising hemoglobin (10.1 g/dL).

On the next day, he was admitted again to the ER for hypotension, melena, and fever. He presented with a hemoglobin level of 8.9 g/dL, leukocytosis, and hyperbilirubinemia. The upper endoscopy revealed several blood clots at the stomach, identifying active hemobilia and an obstructed stent, with no biliary drainage (Figure 1).

A selective common hepatic artery angiography confirmed an arterio-biliary fistula from the proximal right hepatic artery, at the level of a surgical clip (Figure 2). Due to the short length of the fistula, embolization of the fistula tract or the right hepatic artery was not an option. Therefore, placement of a stent-graft was considered the best alternative (Figure 3). Definitive treatment was achieved with the placement of a 5×15 mm stent-graft, in order to preserve arterial liver perfusion.

There was no further bleeding, and the patient was discharged five days later. The patient was followed weekly for a month with complete resolution of the hemobilia and resumption of his daily activities. Although the biliary stent in situ was scheduled for a removal, the patient reported its excretion during a bowel movement. Three years later he remains asymptomatic, with no further episodes of digestive bleeding.

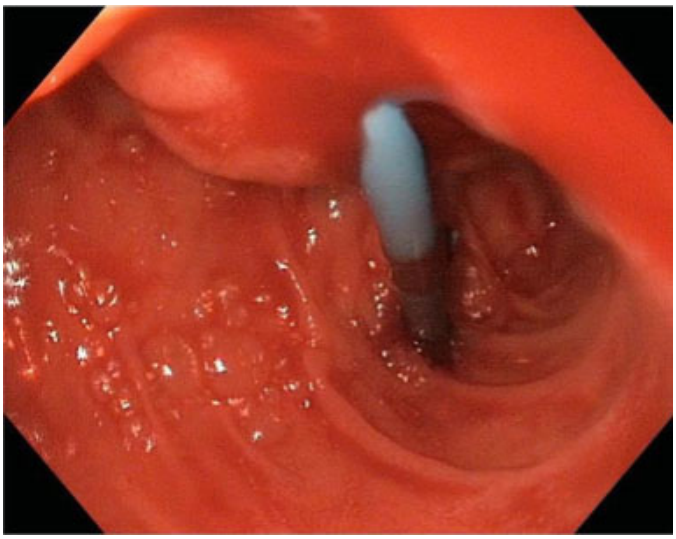

Figure 1: Upper endoscopy image showing an obstructed biliary stent with active hemobilia.

Figure 2: Hepatic angiography. The arrow points to the arterio-biliary fistula in the proximal right hepatic artery.

Figure 3: Hepatic angiography. The bleeding stopped after the placement of the stent-graft. The arrow points to the stent graft in the right hepatic artery.

DISCUSSION

Even though it should be a diagnostic hypothesis in every patient with upper gastrointestinal bleeding after biliary surgery, due to its rarity, hemobilia is usually difficult to diagnose and treat. Misdiagnosis is associated with considerable morbidity and mortality [9].

The most common cause is iatrogenic injury, and, in this case, the main bile duct iatrogenic injury and the required suture might have been the cause for hemobilia. Quincke's triad should be an alert to hemobilia. Usually, there is a two week asymptomatic period before the bleeding starts, but it can be as long as one year [11]. Bleeding may be constant or intermittent; it can be slow and manifest as blood clots in the biliary ducts and melena, or it can be fast and cause hemodynamic instability and hematochezia [3]. Low flow is usually associated with porto-biliary fistula and high flow with arterio-biliary fistula.

The diagnosis is usually made by direct visualization of bleeding from the papilla of Vater through upper endoscopy or ERCP, which may also be therapeutic [2, 12]. Placing a biliary stent or a nasobiliary drain, with or without sphincterotomy, facilitates biliary drainage and leak closure by decreasing the pressure gradient across the sphincter of Oddi [2].

In the present case, the first ERCP did not see active bleeding from the upper biliary tree after clots removal and performed a sphincterotomy. On the following bleeding, the upper endoscopy diagnosed a clot at the sphincterotomy thus, misleading the diagnosis. After confirmation of hemobilia and an arterio-biliary fistula, placing a stent-graft at the right hepatic artery solved the complication. Sphincterotomy could have been avoided if the diagnosis of arterio-biliary fistula had been done at the first ERCP. Some studies even report no advantage on doing sphincterotomy [2].

We decided for conservative treatment with octreotide (to reduce bile secretion) and aminocaproic acid (inhibitor of fibrinolysis), expecting a spontaneous closure of the fistula, which worked only temporarily.

Definitive treatment was achieved with selective angiography, the gold standard to identify and stop the bleeding [10]. Transarterial embolization with coils is preferred over liquid embolic agents because coils cause less distal embolization thus less risk of liver infarction. Clinical success is around 80–100% with low morbidity and mortality rates [3]. If embolization fails or is not feasible, placement of a stent graft can be tried considering the presence of a favorable anatomy. Otherwise, surgical interventions such as vessel ligation or partial hepatectomy are available options, nevertheless with significant risks [3, 13].

CONCLUSION

Physicians should be aware of this uncommon pathology known as hemobilia. Extreme caution must be ensured when doing hepatic resection, and suture of blind areas should be avoided. After surgery, a high suspicion and a swift intervention are necessary to avoid further complications. Definitive treatment is usually achieved with endovascular approaches, thus reinforcing the need for multidisciplinary teams in the management of complex hepatobiliary surgeries.

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Author Contributions

José Pedro Gonçalves – Conception of the work, design of the work, acquisition of data, analysis of data, interpretation of data, drafting the work, revising the work critically for important intellectual content, final approval of the version to be published, agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Pedro Maurício Valente – Acquisition of data, revising the work critically for important intellectual content, final approval of the version to be published, agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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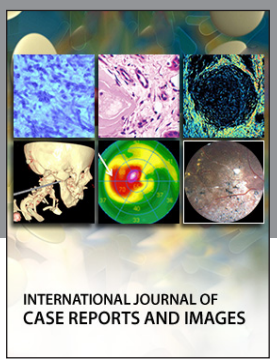

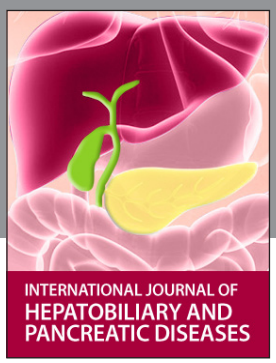

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