CASE REPORT

Venobiliary fistula with haemobilia: a rare complication of percutaneous liver biopsy

Rotimi Ayoola,¹ Parth Jamindar,² Renee Williams³

¹Internal Medicine, New York University Langone Medical Center/Lutheran Medical Center, Brooklyn, New York, USA

²Pulmonary and Critical Care, Augusta University, Augusta, Georgia, USA

³Gastroenterology, New York University Langone Medical Center/Bellevue Hospital Center, New York, New York, USA

Correspondence to
Dr Rotimi Ayoola, rotimi.ayoola@nyumc.org

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SUMMARY

We describe a case of a 36-year-old man with a history of chronic hepatitis C who presented with abdominal pain 3 days after undergoing a percutaneous ultrasound-guided liver biopsy. Initial investigations revealed a venobiliary fistula with haemobilia and obstructive jaundice with direct bilirubin peaking at 9.2 mg/dL. He underwent an endoscopic retrograde cholangiopancreatography with sphincterotomy and stent placement, bilirubin decreased to 3.7 mg/dL, and the patient was discharged. The patient returned with recurrent abdominal pain and upper gastrointestinal bleeding with haemoglobin of 8.6 g/dL, requiring multiple transfusions. He underwent transcatheter arterial embolisation but continued bleeding. Radiofrequency ablation was pursued and was able to achieve haemostasis. The patient was discharged. The patient returned again with a fever of 39.2 °C and was found to have a large right hepatic lobe abscess. The patient underwent abscess drainage with drain placement and was treated with antibiotics for 6 weeks. He followed up in the clinic with resolution of symptoms and infection.

BACKGROUND

Percutaneous ultrasound-guided liver biopsy (PULB) is used to assist in the diagnosis and treatment of liver disease. Venobiliary fistula is a very rare complication of percutaneous liver biopsy leading to haemobilia, bleeding into the biliary tree. Haemobilia caused iatrogenically from percutaneous techniques is the leading cause of haemobilia, while trauma, malignancy, operative injury and inflammatory diseases are other rare causes.¹ In a large series examining percutaneous liver biopsy, the incidence of haemobilia following the procedures was 0.05%.² Many patients remain asymptomatic and achieve spontaneous resolution. However, as this case demonstrates, the presence of signs and symptoms of obstructive jaundice, gastrointestinal bleeding (GIB) and infection, in some patients, poses significant diagnostic and therapeutic challenges, with mortality being reported as high as 25% in a large retrospective series.³

CASE PRESENTATION

A 36-year-old man with a history of gastro-oesophageal reflux disease and chronic hepatitis C presented with abdominal pain. Three days prior, the patient underwent an outpatient PULB. He was not on any regularly prescribed medications and

there was no significant family history. He had a 10 pack-year tobacco history and denied illicit drug use. Vital signs revealed temperature of 36.6°C, heart rate of 66/min, respiratory rate of 17/min, blood pressure of 122/78 mm Hg and oxygen saturation of 100% on room air. Examination revealed a jaundiced-appearing male with normal mental status, scleral icterus, clear lungs bilaterally, normal heart sounds and right upper quadrant (RUQ) abdominal tenderness.

INVESTIGATIONS

Initial complete blood cell count showed haemoglobin level of 14.5 g/dL, white blood cell count of $5.4 \times 10^3/\text{mm}^3$ and platelet count of $182 \times 10^3/\text{mm}^3$. Results of a complete metabolic panel were normal, including a creatinine (0.78 mg/dL), except for an elevated aspartate aminotransferase and alanine aminotransferase of 140 U/L and 145 U/L, respectively, and a total bilirubin of 11.2 mg/dL with direct bilirubin of 9.2 mg/dL. International normalised ratio was 1.2 and lipase was 25 U/L. The rest of his blood tests were unremarkable.

Abdominal CT angiography (CTA) showed a distended gall bladder with intracholecystic blood collection, no evidence of cirrhosis or cholecystitis, and no evidence of blood extravasation from any vessel (figure 1). Hyperbilirubinaemia did not improve, so the patient had a magnetic resonance cholangiopancreatography (MRCP). MRCP was ordered to help identify the source of bleeding and evaluate for common bile duct (CBD) obstruction, arteriovenous fistula and cystic disease, which enabled clinicians to decide which procedure would be most appropriate for the patient, whether it be an interventional radiology procedure, surgical intervention or endoscopic retrograde cholangiopancreatography (ERCP). MRCP revealed intrahepatic and extrahepatic biliary duct dilatation secondary to obstructing haemobilia. The MRCP was also significant for a portal vein to hepatic vein shunt with concomitant fistulous connection to the intrahepatic biliary duct (figure 2).

TREATMENT

ERCP was performed revealing CBD dilatation of 1.5 cm with irregular filling defects. He underwent sphincterotomy revealing copious amounts of blood and fresh clots from the ampulla (figure 3), and a plastic 10 French 7 cm biliary stent was placed (figure 4). Nasobiliary drainage was not considered due to potential patient discomfort.

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Figure 1 Abdominal CT showing a distended gall bladder with intracholecystic blood collection without evidence of cholecystitis.

OUTCOME AND FOLLOW-UP

The patient was hospitalised for 5 days, and his haemoglobin and liver function tests remained stable, direct bilirubin decreased to 3.7 mg/dL, and he was discharged with outpatient follow-up.

The patient returned to the emergency department within a week complaining of worsening RUQ abdominal pain,

Figure 2 Magnetic resonance cholangiopancreatography showing a 1.4x1.3 cm region of hyperenhancement. A branch of the anterior right portal vein (ARPV) leads to this region, while a branch of the middle hepatic vein (MHV) drains this region, revealing a portal vein to hepatic vein shunt with a fistulous connection between the portal vein and hepatic vein shunt and bile duct.

Figure 3 Endoscopic retrograde cholangiopancreatography with 15 mm stone extraction balloon sweep extracting copious amounts of blood and fresh clots.

haematemesis, bright red blood per rectum and melanotic stools. His haemoglobin was 8.6 g/dL and direct bilirubin was 2.6 mg/dL. He underwent blood transfusion. Oesophagogastroduodenoscopy (EGD) showed normal oesophagus, stomach and duodenum without evidence of bleeding. During the EGD the gastroscope was exchanged for a duodenoscope. The stent had remained in place without occlusion or migration, and was

Figure 4 Plastic 10 French 7 cm stent placement.

Figure 5 Abdominal CT showing a radiofrequency ablation with needle tract.

removed. The CBD was then cannulated with a 10mm stone extraction balloon, and the CBD was swept multiple times without the use of fluoroscopy. There was no evidence of blood from the CBD.

CTA showed persistent haemobilia without evidence of active arterial or venous extravasation. Repeat hospitalisation was complicated by persistent haemobilia with recurrent GIB requiring multiple blood transfusions. He initially underwent empiric embolisation to hepatic artery segments 5 and 8. Postembolisation arteriography was performed four times to assess for stenosis in each vessel but could not be confirmed by imaging. Two days after the embolisation, the patient remained anaemic, requiring blood transfusions with continued blood in his stools. The treatment was not fully successful, as the bleeding continued. Haemobilia uncontrolled after embolisation can be due to collateral vessels and may have been the cause of this patient's failure to achieve haemostasis.⁴

It was thought that a broader ablation to the vasculature and possible collaterals would more permanently treat refractory haemobilia. Radiofrequency ablation (RFA) was performed by making a small transverse incision using CT guidance (figure 5). A 4.0cm electrode was then advanced and the tines were deployed to the area of the patient's previous biopsy. Gastrointestinal bleeding resolved after RFA, haemoglobin remained stable and he was discharged after a 17-day hospitalisation.

The patient returned 3 days after discharge with a fever of 39.2°C, and his abdominal CT showed a large right hepatic lobe abscess (8.5×7.2×15.1 cm). The liver abscess was thought to be due to the postprocedural complications of embolisation and/or ablation. The patient was started on intravenous antibiotics with abscess drainage and drain placement. The patient followed up in clinic after 6 weeks of antibiotic treatment with resolution of infection, seen on repeat imaging.

DISCUSSION

Venobiliary fistulas leading to haemobilia can be challenging to diagnose, as most patients are asymptomatic and significant bleeding can occur 3–5 days after the procedure.⁵ Of biliary-vascular fistula cases leading to haemobilia, 40%–60% are iatrogenic, with 38% of those being percutaneous procedures.⁶

Venobiliary fistulas have been associated with surgery, trauma, gallstones, malignancy and percutaneous procedures. In this case a communication between the biliary system and intrahepatic portal venous collaterals was formed by a percutaneous

needle biopsy. However this can be seen in other percutaneous procedures such as transhepatic biliary drainage or transhepatic cholangiography.

Normally, the average hepatic artery pressure is 100 mm Hg. With lower pressures in the CBD (10–15 mm Hg) and portal and hepatic veins (10 and 5 mm Hg, respectively), arterial-biliary fistulas favour blood flow into the biliary system, due to the difference in the pressure gradients.⁷ In contrast, when venous-biliary fistulas form, the pressure gradient is usually too minute for any substantial flow between the venous and biliary systems, with it being uncommon for bile to flow into the bloodstream.⁷

With slower venous flow, the formation of blood clots can form, causing worsening obstruction and jaundice, and may act as a nidus for infection, putting patients at risk for cholecystitis, pancreatitis or cholangitis. Venobiliary fistulas with haemobilia may present with Quincke's triad, which includes epigastric pain, upper GIB and obstructive jaundice; all three may be present in up to 22% of patients.⁶

The choice of diagnostic studies depends on the history, clinical symptoms and level of suspicion. ERCP, EGD, ultrasonography, MRCP and CTA can all be used to aid in the diagnosis. The definitive study of choice is a CTA, which has a 90% detection rate.⁸ Venous fistulas may require a hepatic/late portal phase on CTA or direct portal venography, as these bleeds are slower, which may account for the lack of visualisation on CTA, as in this patient's case. ERCP can be diagnostic in 30% of cases as it allows for direct visualisation of blood from the ampulla of Vater and can be therapeutic in a patient with obstructive symptoms.^{9–12}

The management of haemobilia is aimed at alleviating the biliary obstruction and haemostasis. Conservative management alone often leads to haemostasis. In a review of 171 reported haemobilia cases from 1996 to 1999, 43% of cases were successfully managed conservatively.¹³ However, active interventions may be required when haemostasis cannot be maintained. The mainstays in therapeutic options are transcatheter arterial embolisation (TAE) and surgery. TAE uses selective catheterisation to the bleeding site to stop the blood flow, followed by embolic occlusion, using absorbable gelatin sponge, polyvinyl alcohol particles or microcoils. TAE is the first line of intervention to achieve haemostasis, with a success rate of 80%–100% and lower mortality rates than surgery.⁵ TAE can cause complications including necrosis, hepatic abscess formation as seen in this case, gallbladder fibrosis or arterial thrombosis. Surgery with ligation or resection is reserved for haemodynamically unstable patients, patients with cholecystitis or when TAE has failed, as it has high rates of morbidity and mortality.¹⁴

RFA was successfully used to obtain haemostasis in this patient, but most reported cases of haemobilia detail the use of TAE. RFA's use in the management of haemobilia has not been well described. Kim *et al*¹⁵ report a case of haemobilia secondary to hepatocellular carcinoma refractory to TAE successfully treated with RFA. RFA was able to obliterate a wide vascularity network as well as the small collaterals to ensure haemostasis. RFA can also cause hepatic abscess formation. More studies are needed to validate the use of RFA in the management of haemobilia.

In summary, venobiliary fistulas leading to haemobilia, although uncommon, should be considered in patients presenting with elevated and rapidly progressive serum bilirubin, GIB and fever after a PULB depending on the level of clinical suspicion. If suspected, urgent endoscopy or CTA with late-phase imaging is a very useful diagnostic tool at identifying the cause of bleeding. Conservative therapy and monitoring in haemodynamically

stable patients should be the initial management, as haemobilia is usually self-limiting. Biliary decompression may be needed to restore bile flow. With recurrent bleeding or haemodynamically unstable patients, interventional therapies such as TAE or even surgery may be needed and should be considered in the appropriate setting.⁶

Learning points

- ▶ Venobiliary fistula is a rare cause of haemobilia.
- ▶ Endoscopic retrograde cholangiopancreatography and CT angiography (CTA) are the best investigation techniques to identify haemobilia.
- ▶ Treatment of haemobilia is usually supportive, but if haemodynamic stability or continued bleeding occurs transcatheter arterial embolisation (TAE) or surgery should be considered.
- ▶ Complications of TAE include necrosis, hepatic abscess formation, gallbladder fibrosis or arterial thrombosis.

Contributors Case study written up by RA and PJ, and supervised and edited by RV. RA was clinically involved in the case.

Competing interests None declared.

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