A Rare Case of Fulminant Hemobilia Resulting From Gallstone Erosion of the Right Hepatic Artery

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SUMMARY
Hemobilia is a rare but potentially lethal condition. The commonest cause of hemobilia is trauma, accounting up to 85% of all cases. Hemobilia caused by gallstones is very rare. Most of the cases of hemobilia are either managed conservatively or treated by embolization. Surgery is indicated only when there is an associated surgical condition or when embolization fails. We report a case of a 72-year-old patient with massive hemobilia caused by gallstone erosion to the adjacent artery, diagnosed intraoperatively. The complication was successfully managed by cholecystectomy and repair of the bleeding vessel. This case highlights the importance that hemobilia should be suspected in patients presenting with upper gastrointestinal bleeding. Although rare, massive hemobilia can be life threatening, leading to significant morbidity and mortality. Therefore, a high index of suspicion and timely intervention are important.

INTRODUCTION
Gastrointestinal bleeding from the biliary tree, termed haemobilia, is an uncommon event. A review of haemobilia cases by Sandblom indicated that trauma was the cause of haemobilia in half the cases, with one third of these cases being iatrogenic1. In contrast, non-traumatic haemobilia due to inflammation, gallstone disease and vascular malformations are extremely rare and also difficult to diagnose as compared to traumatic haemobilia. Gallstones accounted for only 5% of all cases2. Here, we present a very rare case of fulminant haemobilia due to gallstone disease, and describe its management.

CASE REPORT
A 72-year-old gentleman was brought to the emergency department after a syncopal attack at home following the passage of large amounts of blackish stools. There was no history of gastrointestinal bleed, abdominal pain or jaundice prior to this. On physical examination, he was pale and jaundiced. No mass was palpable on abdominal examination and per rectal examination revealed fresh melaena. Blood was also aspirated following the insertion of a nasogastric tube. Laboratory tests revealed a haemoglobin of 6g/dL, white cell count of 20,000/mL, total bilirubin of 198 µmol/L with predominant direct hyperbilirubinaemia and serum alkaline phosphotase of 208 U/L. The renal profile and tumour markers were normal.

An oesophago-gastro-duodenoscopy (OGDS) was performed but failed to identify any active bleeding lesion. A side viewing duodenoscopy subsequently revealed intermittent blood and pus oozing from the papilla of Vater. Endoscopic retrograde cholangiopancreatography (ERCP) and common bile duct stenting were carried out. The cholangiogram did not show any filling defect. In view of the finding of haemobilia, an abdominal computerised tomography angiography (CTA) was done. The CTA did not reveal any active contrast extravasation and all visualized vessels were normal in caliber. The gallbladder wall was however thickened, with an irregular space occupying form within the gallbladder lumen suggestive of a gallbladder tumour. A 2.5 x 2.0cm calculus was also seen within the gallbladder. There was pericholecystic fat streaking. No pericholecystic collection was seen. The biliary ducts were not dilated (Figure 1).

In view of persistent ongoing bleeding which required continued blood transfusion, the patient was subjected to surgical intervention with a provisional diagnosis of gallbladder neoplasm. Intra-operatively, a large gallstone was found impacted in Hartmann’s pouch, eroding into the adjacent right hepatic artery. The gallbladder wall was thickened and oedematous with gangrenous change overlying Hartmann’s pouch (Figure 2). Multiple other stones were also found within the gallbladder. Cholecystectomy was performed and the bleeding hepatic artery was repaired with Prolene 6/0 sutures. Haemostasis was secured.

Histopathological examination of the gallbladder showed findings of an empyema gallbladder. Sections of the gallbladder wall revealed diffuse mucosal ulceration and micro-abscess formation extending to the serosa. No evidence of malignancy was identified. The patient was discharged on the eighth post-operative day and remained well on follow up at the outpatient clinic visit 9 months after surgery.

DISCUSSION
Haemobilia is a rare condition that may be difficult to recognize but should be included in the differential diagnoses of upper gastrointestinal bleeding. Haemobilia occurs when there is formation of a fistula between a vessel of the
splanchnic circulation and the intrahepatic or extrahepatic biliary system. Haemobilia in this case was likely caused by pressure necrosis due to an impacted gallstone in the Hartmann’s pouch that gradually eroded into the adjacent right hepatic artery.

Haemobilia associated with gallstones is usually minor, however in this case, the bleeding was so rapid that there was heavy passage of blood into the duodenum, appearing clinically as fresh melena. From a review of published literature, of 13 reported cases of haemobilia caused by gallstones, only three had presented as upper gastrointestinal bleeding. A raised serum bilirubin in this case was most probably due to concurrent Mirrizzi Syndrome, in which the impacted stone in the Hartmann’s pouch was compressing on the common hepatic duct, causing biliary obstruction, or due to blood clots obstructing the bile duct.

In patients presenting with upper gastrointestinal bleeding, OGDS is the first investigation of choice, if available. If blood or a clot is seen at the papilla of Vater, and no other source of bleed is identified, haemobilia is likely to be the cause of the haemorrhage. The choice of subsequent investigation would depend on the history and level of suspicion. Goodnight and Blaisdell in 1981 recommend OGDS, CT and then angiography for cases following trauma.4

The management of haemobilia is directed at stopping the bleeding and relieving biliary obstruction if present. Transarterial embolization (TAE) is now the first line of intervention to arrest bleeding, as the success rate of TAE as reported in most series is 80-100%5. Surgery is indicated when TAE has failed, for cholecystitis, when there is hepatic sepsis and for failure to drain the biliary tree. Haemorrhage from the gallbladder and hemorrhagic cholecystitis require urgent cholecystectomy. As for our patient, CTA failed to identify the site of bleeding and did not detect any vascular abnormality which could likely be treated by embolization. In view of persistent bleeding from an uncertain site and CTA findings suggestive of a gallbladder tumour, emergency surgery was undertaken.

In conclusion, gallstone disease causing erosion of an adjacent artery resulting in active bleeding into the gastrointestinal tract is a rare occurrence, but one which has to be borne in mind when dealing with upper gastrointestinal bleeding in the absence of the more common causes of haemorrhage.

REFERENCES