



A Young Man with Hemobilia Due to a Fistula between Hepatic Artery and Biliary Tract

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ABSTRACT

A 22-year-old man who had suffered two attacks of brisk upper gastrointestinal bleeding (UGIB) within a month prior to admission was diagnosed as having hemobilia because of fistula formation between the hepatic artery and intrahepatic bile duct due to inflammation and abrasion. He was managed by surgery.

KEYWORDS:

Hemobilia, Bile ducts, Inflammation, UGIB.

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INTRODUCTION

The presence of blood in the biliary tree, because of hemorrhage in the biliary tract, has been reported for the first time by Francis Glisson. However, the term hemobilia for the description of this phenomenon was created by Sandblom. Hemobilia is caused by an aberrant connection between the arteries and biliary ducts, which usually presents with one or more of the following symptoms: right upper quadrant (RUQ) pain, upper gastrointestinal bleeding (UGIB), and jaundice.^{1,2}

Usual causes of hemobilia include trauma (accidental and/or iatrogenic), gallstone disease, acalculous cholecystitis, cholangitis, ascariasis or hydatid cyst, hepatic abscess, malignancies of the liver, pancreas and biliary tract polyarteritis nodosa, and vascular malformations of the hepatic artery such as aneurysm or coagulopathy.^{1,3,4,5} Angiography and embolization are the first-line treatment for the management of the fistula between the artery and biliary ducts. Endoscopic Retrograde Cholangiopancreatography (ERCP) and sphincterotomy could be effective in cases of connection between veins and biliary tract. Liver hydatid disease is a rare cause of hemobilia.^{4,6}

Hydatid disease is caused mainly by infection due to cystic or larval stage of the dog tapeworm, *Echinococcus granulosus*. Human, sheep, and other livestock could be intermediate hosts. The disease is more common in rural areas where dogs have access to the infected sheep carcasses. Hydatid cysts are a common cause of conflict in the right lobe of the liver,



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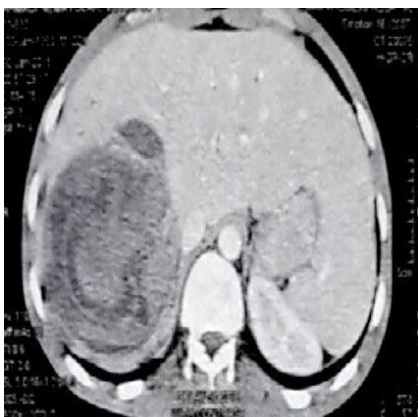


Fig.1: Solid cystic hypodense lesions with septa and calcified areas in the right liver lobe.

especially lower-anterior or lower-posterior segments. Symptoms of the disease include occasional and vague RUQ pain or abdominal distension. Hemobilia is an infrequent cause of UGIB and a rare presentation of hepatic cyst rupture. There are only few case reports in the literature that describes hemobilia due to hepatic cyst rupture.⁴ Both sonography and computed tomography (CT) are sensitive for the diagnosis of hydatid cysts.^{7,8} Surgery is the selective treatment for liver hydatid disease due to the high probability of infection and rupture, unless the cysts are small, or the patient is an inappropriate candidate for surgery.⁹ Albendazole is used before surgery or as a curative approach for small cysts.

CASE REPORT

A 22-year-old man who had suffered two attacks of UGIB within a month prior to admission was referred with symptoms of vague epigastric and RUQ pain without any radiation to other parts of the abdomen. His GIBs had been presented as hematemesis and melena. The patient had no history of heart disease or surgery and had been admitted to hospital, revived, and observed twice in another center. During the previous admission, upper endoscopy showed small size hiatal hernia, gastric antral inflammation, and multiple erosions in gastroesophageal junction (GEJ). Colonoscopy was also normal, while abdominopelvic CT showed solid cystic hypodense lesions with septa and calcified areas with dimensions about $111 \times 110 \times 115$ mm in the right liver lobe (Figure 1) plus mild dilatation of the biliary ducts

and gallbladder sludge, which suggested hepatic hydatid cyst.

At the time of admission, the patient was conscious without any clinical symptoms of GIB. His vital signs were normal, and on examination, his sclera seemed to be mildly icteric, and the conjunctiva was pale. Examination of the neck, chest, and abdomen was normal, and there was not any tenderness, guarding, or distension. His laboratory profile includes Platelet count: 367000, White Blood Cell: $13.8 \times 10^9/L$, Hemoglobin: 7.5 g/dL, Alanine Aminotransferase (ALT): 314 IU/L, Aspartate Aminotransferase (AST): 200 IU/L, Alkaline phosphatase (Alk P): 917 IU/L, Bilirubin (Bili) T: 3.5 mg/dL, D: 1.5 mg/dL, amylase: 41 U/L, lipase: 12 U/L, viral markers negative, and hydatid cyst serology was also negative.

The patient had received Albendazole tablets (400mg twice daily) for 2 weeks. According to the above-mentioned features and diagnosis of hepatic hydatid cyst, he was prepared for surgery. During surgery, the abdomen was opened through a right subcostal incision. A cyst about $100 \times 110 \times 105$ mm was found in the right lobe of the liver within segments 6 and 7. At first, around the cyst and the liver was covered with a sufficient number of gauzes soaked in silver nitrate solution. Then intracystic fluid containing dark blood aspirated by a needle to decompress the cyst. Then, the cyst wall was sectioned and opened, which contained the layers of hydatid cyst, blood, and bile (Figure 2). Cyst contents were evacuated and washed (Figures 3, 4). A small arteriole was found inside the cyst cavity with spurting blood, and a small biliary duct was observed beside the artery and sticking to it with bile leakage, which led to the diagnose of hemobilia (Figure 5). In the next step, hemorrhagic arteriole and biliary duct were ligated. The operation was ended after partial removal of the peri cyst tissue and putting a Jackson-Pratt (JP) -drain inside the cavity.

DISCUSSION

Although triad of hemobilia includes RUQ pain, GIB, and jaundice, most patients do not show all these symptoms.^{1,2} Blood could clot in different places based on the location and amount of bleeding. Hematemesis or melena may occur if the bleeding is brisk or if the blood does not clot in the biliary tract. If blood clots within the

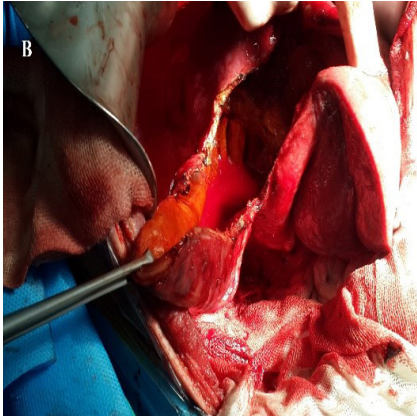


Fig.2: The cyst wall was sectioned and opened, which contained layers of hydatid cyst, blood, and bile.

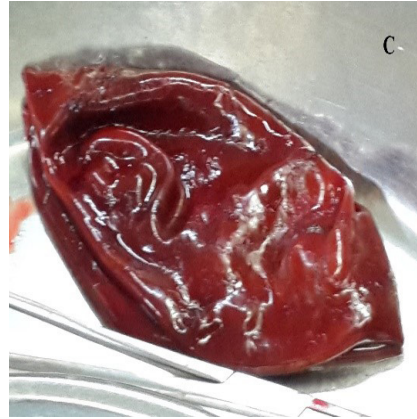


Fig.3: Cyst contents were removed and washed.

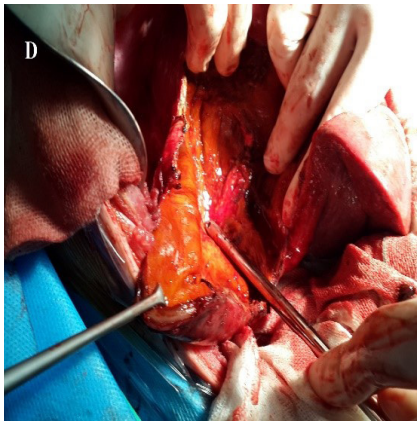


Fig.4: Cyst contents were removed and washed.

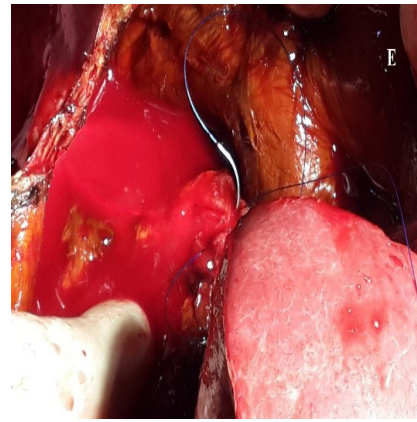


Fig.5: A small arteriole inside the cyst cavity with spurting blood and a small biliary duct sticking to it with bile leakage.

biliary duct, it may cause obstructive jaundice, cholecystitis, or pancreatitis.¹⁰ Our patient presented with upper abdominal discomfort, GIB in the form of hematemesis, melena, and mild jaundice, as well as a slight increase in the diameter of the biliary duct. Raising ALP and bilirubin indicate severe hemorrhage followed by an incomplete obstruction in the biliary tract.

Hemobilia is classified as mild, moderate, or severe, depending on the amount of blood loss and the duration of bleeding. Mild to moderate hemobilia is defined as losing less than 10% of the blood volume, so the patients are hemodynamically stable. Severe hemobilia is diagnosed when hemorrhage is greater than 10% of the total blood volume and results in hemodynamic instability that necessitates transfusion. Our patient is classified as having severe hemobilia due to the large drop of Hb and the need for blood transfusions. An endoscopy can confirm the diagnosis of hemobilia in only 10% of cases by direct

visualization of blood at the ampulla of Vater and is used mostly to rule out other causes of UGIB. ERCP can also be used for diagnosis and is more sensitive in detecting the etiology of hemobilia.¹

Selective angiography can also be applied to confirm the diagnosis of hemobilia, and it could be considered as one of the early measures in cases with severe hemobilia following trauma or known tumors as it is not only diagnostic but is also a therapeutic option. CT can be helpful in detecting hepatic tumors, intraluminal clots, and biliary dilation. It may also demonstrate risk factors related to hemobilia, such as cavitating central lesions and or aneurysms.^{5,8} Magnetic resonance imaging (MRI) can differentiate blood from a stone or sludge and is helpful in suspected cases.⁷ Percutaneous Transhepatic Cholangiography (PTC) and cholangioscopy could also be helpful for the diagnosis of hemobilia. The therapeutic management of hemobilia includes hemostasis,

relieving obstructing symptoms, and replenishing blood loss. Most cases of mild hemobilia could be managed conservatively with the correction of coagulopathy, adequate biliary drainage, and close observation. The first-line treatment for severe hemobilia is transarterial embolization (TAE). Angiography is indicated for severe hemobilia requiring transfusions. Currently, TAE is the gold standard for the treatment of hemobilia, and if unsuccessful, which is rare, surgical treatment is indicated.¹¹ Surgery is usually used for gallbladder or extra-hepatic bleeding, and also in cases of TAE failure or special occasions. Surgery includes direct exploration of the liver with resection risk, ligation, cholecystectomy, and relieving biliary tract obstruction.⁹

While the term hepatic cyst usually means a single non-parasitic cyst of the liver, the cystic lesions of the liver include simple cysts, polycystic liver disease, parasitic, or hydatid cysts, abscess, and or tumor.¹² There are also ductal cysts, choledochal cysts, and Caroli disease, which were not relevant in our patient. Each of these cysts can be diagnosed and cured based on their radiographic appearance and specific features.

In our patient who had hydatid cyst, characteristic daughter cysts were observed within the main cyst. Rupture of cyst did not cause hemobilia in our patient, but arteriolar inflammation and erosion, as well as small intrahepatic bile duct adjacent to hydatid cyst, led to the creation of a fistula between the artery and biliary tract, which were responsible for symptoms that were managed by surgical exploration, excision of hydatid cyst, and ligation of the artery and biliary duct.

CONCLUSION

Hemobilia is a rare cause of UGIB and mostly secondary to trauma and iatrogenic damages. Hepatic cyst, followed by cyst rupture into biliary ducts, is an infrequent cause of hemobilia. In our case, however, the cause of hemobilia was not the cyst rupture, but also the reason was damage to an arteriole and small intrahepatic biliary duct adjacent to hydatid cyst because of the inflammation and formation of a fistula between the artery and biliary duct.

ETHICAL APPROVAL

There is nothing to be declared.

CONFLICT OF INTEREST

The authors declare no conflict of interest related to this work.

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