Hemobilia : Four Case Reports and Review of the Literature

DUANGPORN THONG-NGAM, M.D.*,
PONGSIT WONGKUSOLTHAM, M.D.**,
PINIT KULLAVANIJAYA, MB ChB, DTM&H, FRCP.**
VIBHAKORN SHUSANG, M.D.**,
LINDA BROWN, M.D.***,

Abstract

In this report, we present four cases of hemobilia. Hemobilia occurs when conditions produce an abnormal communication between blood vessels and bile ducts. Although iatrogenic procedures as causes of hemobilia have been reported with increasing frequency, non-iatrogenic etiologies are still quite rare. We, therefore, report 4 cases of hemobilia secondary to different etiologies found in our institution from 1996 to 1998, that are non-iatrogenic. The first patient was a case of congenital aneurysm, the second pseudoaneurysm from trauma, the third cholangiocarcinoma and the fourth hepatocellular carcinoma. The classical triad consists of melena, jaundice and abdominal pain. Direct observation of blood flowing from the Ampulla of Vater by endoscopy was the initial diagnostic procedure in all four cases. Diagnosis was confirmed by ultrasonography, computerized tomography, angiography or surgery. Transcatheter selective embolization as a noninvasive treatment for hepatic aneurysm / pseudoaneurysm is emphasized.

Key word : Hemobilia

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Recognized as a clinical entity in 1654 by Glisson[1], the term hemobilia was first introduced by Sandblom[2] in 1948 to describe the presence of blood in the main biliary tract. The classical presentations consist of colicky abdominal pain, jaundice and melena[3-5]. Hemobilia occurs when disease or trauma produces an abnormal communication between blood vessels and

* Department of Physiology,
** Department of Medicine, Gastroenterology Unit,
*** Department of Radiology, Faculty of Medicine, Chulalongkorn University, Bangkok 10330, Thailand.

bile ducts. This is called arteriobilous fistulae[6]. Once considered a rare condition, hemobilia is now being reported with increasing frequency. Hepatic causes currently represent approximately 50 per cent of cases and may be of trauma, vascular [7-9], inflammatory[10-12], infectious[13] or neoplastic[14,15] in etiologies. Several cases of hemobilia have been reported in patients with bleeding tendency such as idiopathic thrombocytopenic purpura[16]. Reports from three large series reveal that trauma is the leading etiology of hemobilia (Table 1). The rising most likely reflects the increased use of percutaneous liver biopsy[17-19] ; complications following laparoscopic cholecystectomy[20], percutaneous biliary drainage and post sphincterotomy[21,22,26].

CASE REPORT

Case 1.

A 19-year-old Thai man was admitted after 1 day of shaking chills, vomiting, jaundice, right upper quadrant abdominal pain, and melena. His past medical history was unremarkable. The patient denied any prior hepatobiliary symptom, nor any history of alcohol abuse, overseas travel, abdominal trauma, or peptic ulcer disease.

On admission, the patient was found to have a temperature of 102°F and a blood pressure of 110/80 mmHg without orthostatic changes. Physical findings were jaundice and right upper quadrant tenderness on deep palpation of the abdomen. The patient's stool was positive for occult blood. The initial hematocrit was 43 per cent and the white blood cell count was 14,400/ mm^3 with 95 per cent segmented neutrophils. Clotting parameters were within normal limits. Liver function test showed 18 mg per cent total bilirubin with 5 mg as direct fraction, alkaline phosphatase 219 units (normal, ≤ 279), aspartate aminotransferase 167 units (normal, ≤ 38), alanine aminotransferase 12 units (normal, ≤ 38) and serum albumin 3.5 mg/ml. Chest and abdominal roentgenograms were within normal limits.

On the first hospital day intravenous fluid was instituted and broad-spectrum antibiotics were administered after blood cultures had been obtained. Ultrasonography of the abdomen showed dilatation of intrahepatic bile ducts and common bile duct containing echogenic material. Computed Tomography of the abdomen confirmed dilatation of intra-and extra-hepatic bile ducts. Hypodense contrast was noted filling within the dilated duct (Fig. 1). These findings were consistent with either blood clot or muddy stones as a cause of bile duct obstruction.

On the following day, the patient underwent arteriogram. Selective coeliac and hepatic arteriography showed a small aneurysm of the branch of the left hepatic artery, (Fig. 2). The embolization was performed using gel foam. Selective hepatic arteriography, post embolization, showed complete resolution of the aneurysm (Fig. 3).

After embolization procedure, the patient was stable with no evidence of gastrointestinal blood loss. He recovered dramatically and finally was discharged.

Case 2.

A 47-year-old woman was referred from another hospital because of melena for one day. Thirteen days previously she had blunt trauma of the abdomen from a car accident. After that, she had biliary colic attack and progressive

Table 1. Hemobilia cases : number (percentage) by causes.

	Sandblom (1972) (n = 355)	Curet (1984) (n = 86)	Yoshida (1987) (n = 103)
Trauma (iatrogenic)	59 (17)	50 (58)	42 (41)
(accidental)	137 (38)	23 (27)	20 (19)
Gallstone-related	53 (15)	-	9 (9)
Inflammatory	46 (13)	-	10 (10)
Primary vascular	38 (11)	-	15 (14)
Neoplastic	22 (6)	-	7 (7)
Other	-	13 (15)	-

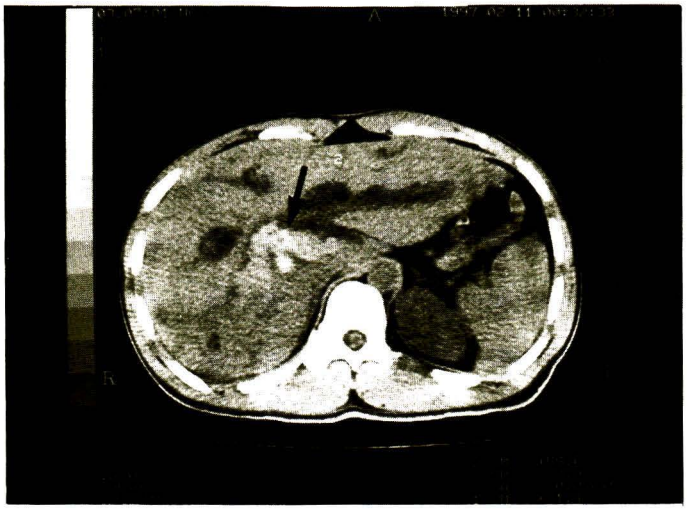

Fig. 1. CT scan show dilated intrahepatic bile ducts containing hyperdensity lesions, representing blood clots (arrows).

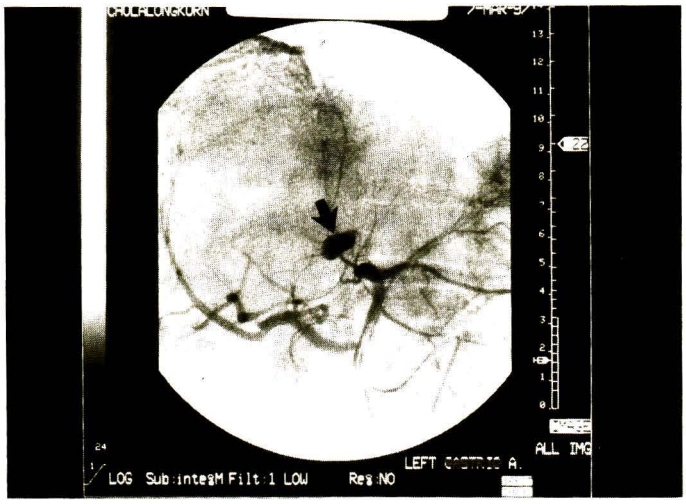

Fig. 2. Angiography shows small aneurysm from a branch of left hepatic artery (arrow).

jaundice of which the former slowly improved and was gone in eight days. CT scan of the upper abdomen from another hospital showed hepatic laceration with intrahepatic and subcapsular haemorrhage.

Three days before referral, she had another biliary colic attack and then developed melena. Upper GI endoscopy was performed and revealed coffee ground content in the second part of the duodenum. She was then referred to our hospital.

Physical examination showed stable vital signs, mild pallor, moderate jaundice, with melena on digital rectal examination. The initial hema-

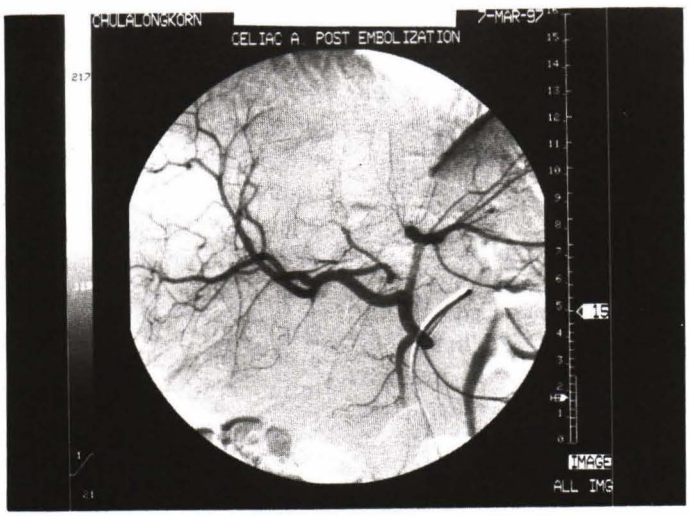

Fig. 3. After embolization with gel foam, the aneurysm has disappeared, indicative of successful treatment.

tocrit was 29 per cent and the white blood cell count was 10,580/mm^3 with 72 per cent segmented neutrophils. Liver function test showed 10 mg per cent total bilirubin with 7 mg as direct fraction, alkaline phosphatase 1,242 units (normal, ≤ 279), aspartate aminotransferase 347 units (normal, ≤ 38), alanine aminotransferase 711 units (normal, ≤ 38) and serum albumin 3.9 mg/ml.

Upper GI endoscopy revealed blood oozing from the Ampulla of Vater. Angiogram showed hepatic arterial pseudoaneurysm which was embolized successfully. The patient made a full recovery.

Case 3.

A 59-year-old monk from the north eastern part of Thailand presented with jaundice, right upper quadrant (RUQ) abdominal discomfort, anorexia and weight loss for 1 month. During admission he developed RUQ abdominal pain and melena, of which upper GI endoscopy revealed blood oozing from Ampulla of Vater. CT scan of the upper abdomen showed a 2-cm mass at the porta hepatis with diffuse dilatation of intrahepatic bile ducts and paraaortic lymphadenopathy. Angiography was performed but no active bleeding was detected. At laparotomy, a mass at the common hepatic duct was found and T-tube choledochostomy was performed for palliative treatment of biliary obstruction. Biopsy of the mass proved to be cholangiocarcinoma.

Case 4.

A 57 year-old-man who had cryptogenic cirrhosis as an underlying disease and had undergone a hepatectomy three years previously due to hepatocellular carcinoma (HCC), had normal ultrasonography of hepatobiliary system and serum α-fetoprotein measurement during follow-up. Three days prior to admission, the patient developed melena and orthostatic hypotension. Upper GI endoscopy was done and active bleeding from Ampulla of Vater was detected. Bleeding spontaneously stopped without any intervention. Computerized tomography with contrast study showed cirrhotic liver and splenomegaly. Hypodense area with heterogenous enhancement at the right posterior aspect of the remaining liver was observed and the provincial diagnosis was recurrent HCC. Liver biopsy was contraindicated due to coagulopathy and severe thrombocytopenia. We, then, proceeded to do transcatheterized oily chemoembolization (TOCE) for pallative treatment of HCC.

DISCUSSION

The clinical features of hemobilia include colicky abdominal pain, jaundice and gastroin-

testinal bleeding[3-5]. The bleeding may range from scant[21] to profuse and intermittent to continuous, and may arise from communication to the biliary tree at any level. Blood clots in the biliary system may cause acute biliary obstruction which leads to the development of cholangitis, cholecystitis[19], or pancreatitis[3], or the clot may serve as a nidus for biliary calculi.

The age distribution of patients with hemobilia tends to peak in the sixth decade of life, representing the amount of iatrogenic trauma in people in this age group undergoing invasive testing and treatment. A second concentration of patients occurs in the first four decades of life, representing the higher likelihood of accidental trauma in this age group.

The first two patients were cases of hemobilia that were caused definitely by congenital aneurysm and pseudoaneurysm respectively. The third cholangiocarcinoma and the fourth hepatocellular carcinoma were assumed to be caused by hemobilia by tumoral ductal migration[28].

The approach to the management of hemobilia involves initial resuscitation and evaluation, including laboratory analysis of hematocrit, coagulation, renal and liver functions. When hemorrhage into the biliary tract is suspected, the following diagnostic procedures should be undertaken. The first measure is gastrointestinal endoscopy to rule out other bleeding sources. Direct observation of blood flowing from the Ampulla of Vater is not uncommon in hemobilia. Endoscopy may be combined with retrograde cholangiopancreatography, which sometimes reveals clots in the ducts.

Ultrasonography or computed tomography often demonstrates even small traumatic lesions. The sonographic appearances of intrahepatic blood vary and include biliary dilatation with echoic material visible in the extrahepatic bile ducts[23]. Clotted blood in the biliary tract produces less distinctive shadows than stones. CT scan may show a hyperdense blood clot within dilated bile ducts, as seen in our first case.

The best way to verify the diagnosis is by selective arteriography, which reveals the source of bleeding in a high percentage of cases by displacement of the vessels around a liver mass or by filling of an aneurysm[24]. Selective embolization can cure the cause of hemorrhage and prevent further complications from surgical approach in some cases.

Treatment decision for hemobilia should be based on the anatomical source of bleeding. In the past, the treatment for intrahepatic causes of hemobilia was ligation of the main hepatic arterial branch. Now the ability to perform subselective embolization has improved greatly[25-27]. The complication rates of transcatheter embolization are low. This low trauma method allowed either a complete control of bleeding or help reduce intraoperative blood loss by more than half. Since 20 per cent of patients develop minor liver damage, manifested as transient elevation in serum liver enzymes levels, these values should be monitored after the procedure. If highly selective embolization is unsuccessful, embolization of the main hepatic artery or its major branches can be performed. Surgical treatment is also very successful if transcatheter therapy fails[17]. However, as initial therapy, surgery does not approach the cure rate and low morbidity of transcatheter embolization. Surgical therapy includes liver suturing, partial hepatic resection, and vessel ligation.

When hemobilia accompanies major liver injury, initial surgery with debridement, vessel ligation, and drainage is important, but transcatheter therapy should also be considered as an adjunct to control vascular injuries. Rare portal venous causes of hemobilia should be treated surgically. Surgical therapy has a reported 77 per cent success rate in extrahepatic causes of hemobilia. Cholecystectomy is the procedure of choice in cases where the gallbladder is the source of bleeding. Subselective embolization has been reported for massive cystic artery hemorrhage, but adjunctive cholecystectomy should be planned. Resectable neoplasms and common bile duct tumors should also be approached surgically. Embolization may be considered for unresectable bleeding lesions if technically possible. Expectant observation is an option for managing hemobilia in selected cases. Spontaneous cessation of bleeding will occur in one third of patients with hemobilia after percutaneous liver biopsy. Some authors have recommended the placement of clot promoters into percutaneous needle puncture tracts to reduce bleeding complications.

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ภาวะเลือดออกจากท่อทางเดินน้ำดี รายงานผู้ป่วย 4 ราย และทบทวนวรรณกรรม

ดวงพร ทองงาม, พ.บ., วิภากร ชูแสง, พ.บ.**, พงษ์สิทธิ์ วงศ์กุศลธรรม, พ.บ.**, ลินดา บราวน์, พ.บ.***, พินิจ กุลละวณิชย์, พ.บ.***

ผู้รายงานได้นำเสนอผู้ป่วย 4 ราย ที่มีภาวะเลือดออกจากท่อทางเดินน้ำดี (hemobilia) ซึ่งเป็นภาวะที่พบได้ไม่บ่อย เกิดได้จากการมีทางเชื่อมระหว่างเส้นเลือดกับระบบทางเดินน้ำดีโดยมีสาเหตุมาจากความผิดปกติของผู้ป่วยเอง หรือโรคจากแพทย์ทำ ในรายงานนี้พบสาเหตุของ hemobilia จากความผิดปกติของเส้นเลือดที่โป่งพอง (true และ pseudoaneurysm) มะเร็งท่อทางเดินน้ำดี (cholangiocarcinoma) และมะเร็งตับ (hepatocellular carcinoma) อาการหลัก 3 ประการ ที่พบคือ เลือดออกจากทางเดินอาหารส่วนต้นโดยอาเจียนเป็นเลือดหรือถ่ายดำ ตัวเหลืองตาเหลือง และปวดท้องด้านขวาบน การวินิจฉัยเริ่มต้นจากการพบเลือดออกจากรูเปิดทางเดินน้ำดี (ampulla of Vater) โดยการส่องกล้องตรวจทางเดินอาหาร ตรวจยืนยันได้จากอุลตราซาวด์ เอกซเรย์คอมพิวเตอร์ หรือฉีดสีเพื่อดูระบบทางเดินน้ำดี ส่วนการฉีดสีเส้นเลือดแดงใช้ในการวินิจฉัย และรักษาโดยใช้สารเจลโฟมอุดตันเส้นเลือดที่ผิดปกติพบว่าได้ผลดี

คำสำคัญ : Hemobilia

ดวงพร ทองงาม, วิภากร ชูแสง, พงษ์สิทธิ์ วงศ์กุศลธรรม, ลินดา บราวน์, พินิจ กุลละวณิชย์
จดหมายเหตุทางแพทย์ ฯ 2544; 84: 438–444

* ภาควิชาสรีรวิทยา,
** ภาควิชาอายุรศาสตร์, หน่วยโรคระบบทางเดินอาหาร,
*** ภาควิชารังสีวิทยา, คณะแพทยศาสตร์ จุฬาลงกรณ์มหาวิทยาลัย, กรุงเทพฯ 10330