

## Haemobilia - A Rare Cause of Upper Gastro-Intestinal Bleeding

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### Rezumat

#### *Haemobilia – o cauză rară de hemoragie digestivă superioară*

Haemobilia reprezintă o cauză rară de hemoragie digestivă superioară ce se manifestă la nivelul arborelui biliar. Majoritatea cazurilor sunt de cauză iatrogenă, colecistectomia laparoscopică sau clasică, traumatismele abdominale, litiaza căii biliare principale, tumori hepatice, aneurisme. Prezentăm cazul unui pacient internat în Clinica de Chirurgie pentru dureri în epigastru și hipocondrul drept, greață și vărsături. S-a practicat colecistectomie clasică și drenaj biliar extern pe tub transcistic. Evoluția postoperatorie a fost favorabilă cu scăderea continuă a valorilor hemoglobinei. În ziua 13 post-operator drenajul biliar a fost de 800 ml – aspect de biliragie. Starea generală a pacientului s-a alterat, cu apariția melenei și a hematocheziei. S-a practicat endoscopie, colangiografie și examen computer tomografic abdominal. Episodul s-a repetat în ziua 27 postoperator. S-a practicat duodenotomie și explorarea arborelui biliar. Angiografia din ziua următoare a evidențiat fistula arterio-biliară în segmentul IV hepatic, urmată de embolizare. Haemobilia a reapărut 15 zile mai târziu când s-a practicat colonoscopie și angiografie cu embolizare cu spirale metalice. Evoluția pacientului a fost favorabilă, externându-se la 13 zile de la embolizare. Angiografia intervențională rămâne opțiunea terapeutică în cazul hemobiliei. Alte opțiuni precum ligatura arterei hepatice sau hepatec-

tomia reglată reprezintă alternative în cazul ineficienței embolizării arteriale.

**Cuvinte cheie:** hemobilie, angiografie, colecistectomie

### Abstract

Haemobilia is a rare cause of upper gastrointestinal bleeding that consists of haemorrhage within the biliary tree. Most cases of haemobilia are due to iatrogenic cause, laparoscopic or open cholecystectomy, abdominal trauma, gallstones, hepatic tumours, vascular aneurism. We present the case of a male patient admitted in the surgery department for epigastric and right hypochondria pain, nausea and vomiting. Open cholecystectomy was performed with a trans-cystic tube drainage. Postoperative outcome was favourable but with a continuous decrease in haemoglobin level. In the 13<sup>th</sup> day postoperatively biliary drainage was 800 ml - haemobilia. Patient health status altered and melena and hematemesis occurred. Endoscopy, cholangiography and abdominal computer tomography (CT) were performed. The episode repeated in day 27 after initial surgery. Duodenotomy and exploration of the biliary tree was performed. Angiography was performed next day that revealed biliary-arterial fistula within segment IV of the liver followed by embolization. Haemobilia reoccurred fifteen days later and colonoscopy and angiography were performed. Embolization with metallic coils was performed. Patient outcome was favourable and was discharged 13 days after second embolization. Interventional angiography remains the first treatment option of haemobilia. Selective arterial ligation or hepatectomy remain the options in case of lack of angiography or insufficient results after embolization.

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**Key words:** haemobilia, angiography, cholecystectomy

## Introduction

Haemobilia is a rare cause of upper gastro-intestinal bleeding that consists of a hemorrhage within the biliary tree. It is often due to a complex pathology, difficult to diagnose and treat.

Classically it is associated with Quincke triad (biliary colic, jaundice and upper gastro-intestinal bleeding). It occurs in only 40% of cases. In the beginning, the most important signs and symptoms are shadowed by others, that make the diagnosis even more difficult. Most cases of haemobilia are due to iatrogenic cause, laparoscopic or open cholecystectomy being one of them. Other causes of haemobilia include abdominal trauma, gallstones, hepatic tumors, vascular aneurism. Arterial-biliary fistulae is a rare cause of haemobilia.

## Case report

We present the case of a male patient, aged 58 years, from the county-side that was admitted to General Surgery Department for epigastric and right hypochondria pain, nausea and vomiting.

Patient history showed: diabetes mellitus controlled with oral medication, severe right lower limb arteriopathy due to diabetes mellitus, dyslipidemia, blood hypertension, ischemic cardiomyopathy. The patient was under medication for metabolic and cardiologic disorders.

Clinical examination revealed: distended abdominal wall, mobile, sensitive in upper quadrants with positive Murphy maneuver. There was a postoperative wound dehiscence after a right toe resection performed two weeks before in the Cardiovascular Surgery Department.

Blood samples at admission showed: leucocytes 23590 / mm<sup>3</sup>, hemoglobin 10.5 g/l, thrombocytes 534 000 / mm<sup>3</sup>, glucose 264 mg/dl, total bilirubin 3.79 mg/dl, direct bilirubin 3.34 g/dl, AST 747 U/l, ALT 837 U/l, serum amylase 2866 U/l, serum lipase 6937 U/l.

Abdominal sonography revealed: thickened walls of gallbladder, with sludge (50%), non-dilated intrahepatic biliary tree, main bile duct 6 mm, normal pancreas, no fluid in the peritoneal cavity.

Computer tomography (CT): increased size liver; cranial-caudal diameter of right hepatic lobe = 190 mm, with diffused lower density, dilated intrahepatic biliary tree, especially closer to hepatic hilum, dilated main bile duct – 12 mm at hepatic hilum, 9 mm in the retropancreatic segment. 3 mm gallstone within main bile duct. Gallbladder of 90/50 mm, with thin walls, and dense bile. A mixed gallstone in the infundibulum. Increased sized pancreas, with the head of 38 mm, diffuse margins. Diffuse increased density of peri-

pancreatic fat tissue, especially around pancreatic head and root of mesentery. Visible Wirsung all along with a maximum diameter of 3.9 mm. Normal CT aspect of spleen, kidneys, adrenal glands. No visible fluid in the peritoneal or pleural cavities.

After initial resuscitation, open cholecystectomy was performed with a trans-cystic duct drainage. During surgery a dilated gallbladder was observed, thickened walls, a thin cystic duct and inflammatory process within the hepatic pedicle. There was also fat necrosis at the level of transverse colon.

Postoperative outcome was favorable but with a continuous decrease in hemoglobin level until 5.7 g/dl in the 13<sup>th</sup> day postoperatively. Biliary drainage in the same day was 800 ml – haemobilia. Patient health status altered and melena and hematemesis occurred.

Emergency endoscopy was performed and revealed normal stomach, duodenum with fresh blood in large quantity.

Colangiography revealed heterogeneous aspect of a branch of right hepatic duct, normal aspect of Wirsung and no image of the gallstone within main bile duct.

CT examination revealed: trans-cystic drainage tube within the right hepatic duct, sub-hepatic drainage tube in proximity of hepatic hilum, pneumobilia within right intrahepatic ducts, non-dilated intrahepatic and extrahepatic bile ducts, small blood density within distal choledocus. Fluid density at hepatic hilum, along the visceral surface of caudate lobe until the right diaphragmatic crus, along the common hepatic artery, along portal vein, along head and body of pancreas, along descending duodenum. Inferior limit of the liver situated below ribcage, relatively homogenous pancreas, normal spleen, kidneys, adrenal glands. Pericardic fluid with 1cm thickness.

In the 27<sup>th</sup> day postoperatively, due to lack of angiography, during night, exploratory laparotomy was performed, gastrotomy, duodenotomy and exploration of greater duodenal papilla – bleeding at its level was observed. Catheterization of the papilla was performed and lavage with serum. Bile and blood / cloth were evacuated. No tumor was detected at the level of pancreas, duodenum, bile duct.

Angiography was performed in the 28<sup>th</sup> day postoperatively through femoral approach. It revealed leakage from a small branch of hepatic artery that supply segment IV. Gelaspon embolization was performed with optimum result. (Fig. 1)

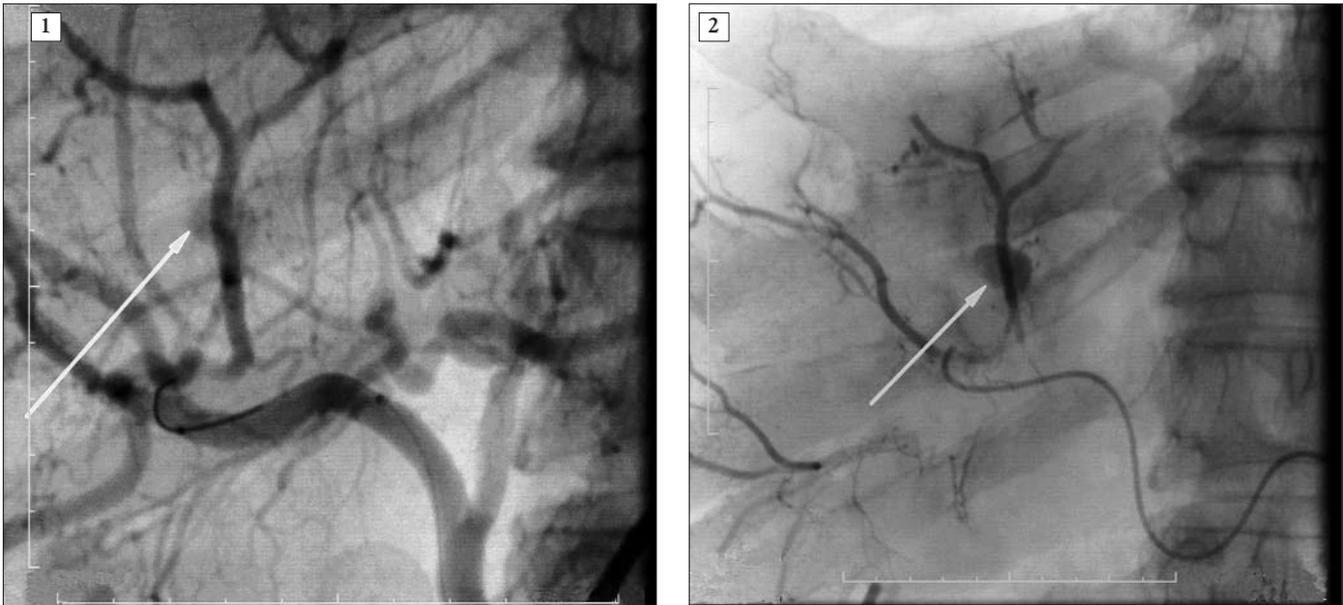
In the 7<sup>th</sup> day after angiography (35<sup>th</sup> day postoperatively) patient is stable with increase values of hemoglobin. Then in the 15<sup>th</sup> day after embolization, the patient becomes unstable, with severe anemia and hematochezia (Hgb 5g/dl).

Colonoscopy shows blood in large quantity inside colon without an active bleeding source.

Angiography shows same biliary-arterial fistulae. Embolization with two metallic coils for cerebral aneurism was performed. (Fig. 2)

Post-angiography evolution is favorable with normalization of blood samples values. The patient was discharged in day 57 after admission (13 days after last embolization).

The patient came for medical check-ups for the next 60 days. No bleeding signs occurred in this period. (Fig. 3)



Figures 1,2. Angiography

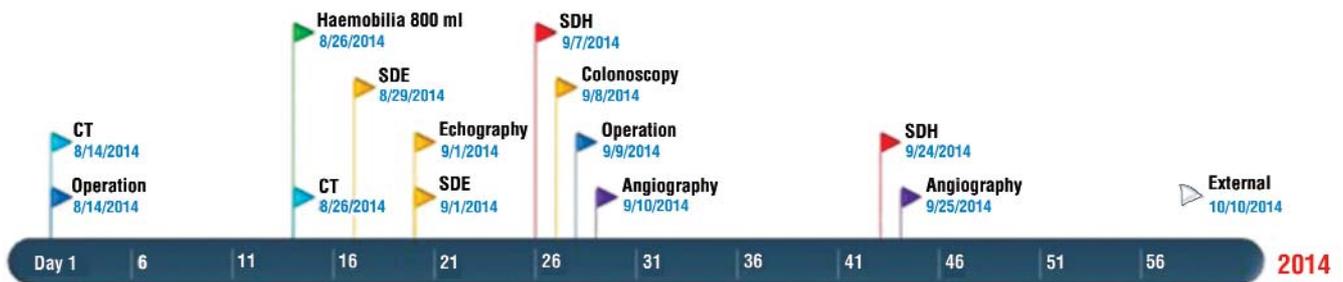


Figure 3. Evolution of patient events during hospital stay

**Discussions**

Initially the main cause for haemobilia was trauma. Recent studies change this and most reported cases are iatrogenic. Due to its rarity and difficult diagnosis a guideline is difficult to be established. (1,2)

Among the causes of iatrogenic haemobilia we can list recent or late laparoscopic or classic cholecystectomy, Whipple procedure, cystic or hepatic artery aneurism, liver or renal biopsy puncture or biliary stenting. (2-7)

Decrease of hemoglobin that associates with increase values of total bilirubin and direct bilirubin may suggest haemobilia. Furthermore, increase values of AST/ALT, GGT and amylase and lipase and jaundice come to underline haemobilia. (7-9)

Increased values of amylase, lipase and jaundice may be due to irritation of Oddi sphincter or due to its obstruction by a cloth.

In this case, the initial image of a gallstone within bile duct (initially seen in CT exam and not confirmed with sonography) may underline the cloth hypothesis. This theory is sustained by the imaging aspect of main bile duct and lack of gallstones.

The initial moderate anemia at admission may be due to diabetes mellitus nephropathy or as a pre-existence of the biliary-arterial fistulae in segment IV (that was diagnosed in the further days).

Haemobilia is a rare clinical entity. The physician should have a complete image of all clinical and imaging aspects of the case that should lead to a rapid indication for interventional angiography. (10,11)

**Conclusions**

Although it is a rare pathology haemobilia must not be a deferral diagnosis.

Treatment of this pathology needs a specific infrastructure,

a mixed and well trained team.

In case of absence of any of these factors, the patient must be guided towards a specialized medical center.

Interventional angiography remains the first treatment option of haemobilia. Selective arterial ligation or hepatectomy remain the options in case of lack of angiography or insufficient results after embolization.

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