

Case Report

Hemobilia, an uncommon case in a patient with severe acute cholangitis: a case report

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Received: 29 October 2024

Accepted: 07 December 2024

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ABSTRACT

Hemobilia is an uncommon cause of upper gastrointestinal bleeding and it is usually associated with hepatobiliary trauma. We present a 71-year-old female patient with type 2 diabetes mellitus and hypertension, who was admitted to the emergency department due to clinical features of a severe acute cholangitis and hemodynamic instability. She required an external drainage through a T-tube in the common bile duct and admission to the intensive care unit in order to improve clinical conditions for a definitive treatment. Afterwards we performed an open cholecystectomy with a satisfactory postsurgical outcome and decided to discharge her. However, she was readmitted due to melena and bleeding through the T-tube, which developed a hypovolemic shock treated with crystalloid fluids and blood transfusion. It is still in discussion what is the best treatment option. Since the objective is to preserve the integrity of the bile duct, only 5% of the cases will require a major intervention. It is important to individualize every case.

Keywords: Hemobilia, Cholangitis, Case report, ERCP, Bile ducts

INTRODUCTION

Hemobilia is defined as a bleeding from or into the biliary tract, which was first described by Sandblom in a patient with biliary trauma in 1948.¹ It is an uncommon cause of digestive bleeding and usually manifests with hematemesis or melena. According to a case series of 222 patients by Takehiro Abiko et al, the most common etiologies were iatrogenic (65%), inflammatory (13%), neoplastic (7%), traumatic (6%) and others (9%).²

The typical clinical features reported by Quincke in 1871 were biliary colic, jaundice and upper gastrointestinal bleeding, which are present only in the 22% of the patients.³ Treatment depends on the presentation and it is still in discussion, given that it is relevant to keep the integrity of the bile duct. Therefore, the gold standard is the hepatic artery embolization.

CASE REPORT

A 70-year-old female was admitted to the Emergency Department due to pain in the upper right abdomen for 72 hours associated with jaundice, fever and nausea. She had a history of hypertension and type 2 diabetes mellitus for 20 years and had been taking losartan and metformin. Her past surgeries were caesarea and total abdominal hysterectomy due to uterine fibroids. Physical examination revealed evident jaundice, decreased bowel sounds and Murphy's sign was positive. Her mean blood pressure was less than 65 mmHg, for which vasopressor medication was indicated. Blood tests showed mild anemia, hyperglycaemia, leukocytosis, elevated liver enzymes with a cholestatic pattern, and mild hypokalemia (Table 1). Liver ultrasound revealed a 133×39×59 mm gallbladder with 4 mm wall thickness, a 13 mm calculus at the gallbladder's neck, dilatation of 14 mm of the common bile

duct and positive sonographic murphy sign. Based on these findings, the diagnosis of severe acute cholangitis was made. Therefore, an external biliary drainage was performed, and piperacillin/tazobactam was prescribed postoperatively with a satisfactory outcome.



Figure 1: Bleeding from the Kehr tube.

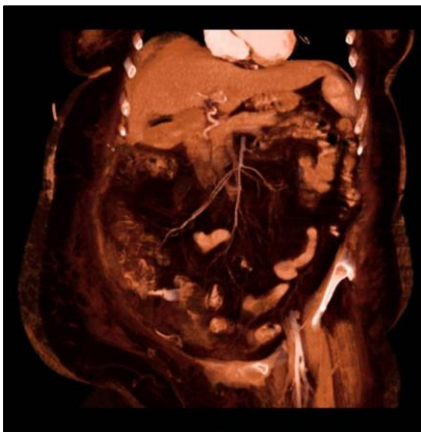


Figure 2: Angiotomography without evidence of arterio-biliary fistula.

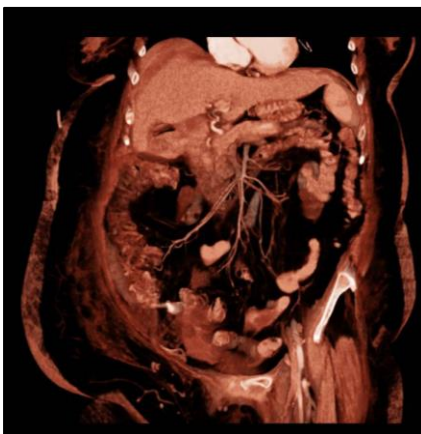


Figure 3: Angiotomography without evidence of pseudoaneurysm.

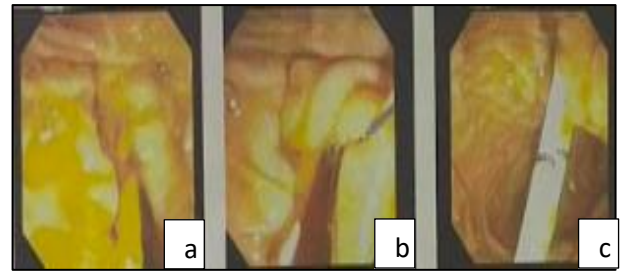


Figure 4 (a-c): Cholangitis associated with discharge of purulent liquid and presence of a 12×8.5 cm functional biliary prosthesis.

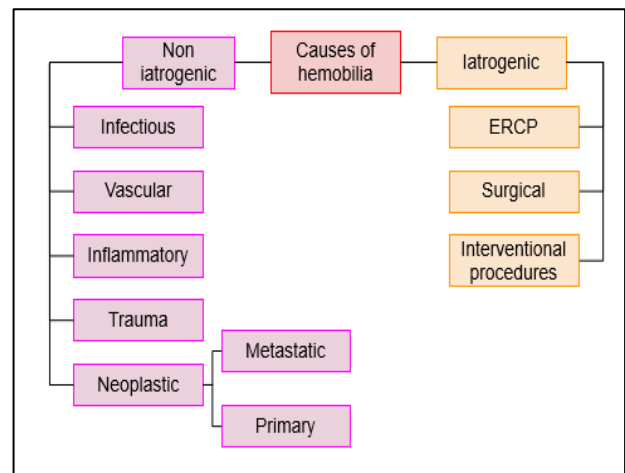


Figure 5: Causes of hemobilia.

On the fifth day, she underwent an open cholecystectomy with exploration of the common bile duct, which revealed the following findings: dilated bile measuring approximately 15 mm, multiple gallstones, biliary sludge and abundant purulent fluid. Intraoperative blood loss was 400 cc.

She was admitted to ward for medical management and was discharged on the 8th day due to clinical improvement. Seven days after discharge, she returned with bleeding from the Kehr tube (Figure 1), hematemesis and melena, resulting in hypovolemic shock. Blood tests on readmission revealed severe anemia and leukocytosis (Table 1).

Authors initiated fluid resuscitation with crystalloids and blood transfusion. She showed partial recovery and spontaneous control of the bleeding. During hospitalization, she developed atrial fibrillation exacerbated by metabolic stress, which was treated with an infusion of amiodarone and improved alongside the control of acute bleeding.

A follow-up ultrasound and an angio tomography did not report any vascular injury, fistula or pseudoaneurysm (Figures 2 and 3). She presented 3 episodes of self-limited bleeding. However, during the last one we performed an

endoscopic retrograde cholangiopancreatography (Figure 4), removed the external biliary drainage and placed a biliary stent. She developed mild acute post-ERCP pancreatitis which resolved 48 hours later. Discharge was

on the 15th day due to clinical improvement and had follow-up by the surgery department without any new episode of bleeding.

Table 1: Laboratory findings.

Laboratory tests	Admission	Readmission	Post-ERCP	Normal range
Haemoglobin	11.7 g/dl	6.0 g/dl	8.5 g/dl	12.50-16.80
Hematocrit	35%	17.8%	24.9	36-47
Platelets	154×10 ³ µl	286 ×10 ³ µl	194×10 ³ µl	142-424
Leukocytes	19.6 ×10 ³ µl	18.30×10 ³ µl	6.40×10 ³ µl	4.07-11.23
Glucose	201 mg/dl	150.3 mg/dl	92.7 mg/dl	74-106
BUN	20 mg/dl	26.5 mg/dl	-	7-25
Urea	43 mg/dl	56.7 mg/dl	14.6	17-43
Creatinina	1 mg/dl	1.3 mg/dl	0.4 mg/dl	0.66-1.09
Total bilirubin	7.4 mg/dl	-	2.1 mg/dl	0.30-1.20
Indirect	2.9 mg/dl	-	-	-
Direct	4.5 mg/gl	-	1.1 mg/dl	-
AST	92 u/l	-	20 u/l	0-35
ALT	79 u/l	-	13 u/l	0-35
LDH	172 u/l	-	-	140-271
GGT	601 u/l	-	95 u/l	9-64
Sodium	142 mEq/l	-	140.2 mEq/l	136-145
Potassium	3.2 mEq/l	-	3.4 mEq/l	3.50-5.10
Chloride	107 mEq/l	-	111 mEq/l	98-107
Prothrombin time	13.1	16	15.1	9.10-12.10
INR	1	-	1.31	0.80-1.10
Partial thromboplastin time	26.8	37.8	30.2	< 40
Amylase	-	-	158 U/l	22-80
Lipase	-	-	501 U/l	0-67

DISCUSSION

Non-variceal upper gastrointestinal bleeding is still a diagnostic and therapeutic challenge, representing a mortality rate of 10 - 36%. The most common causes involve acid peptic disease, Mallory-Weiss syndrome, erosive gastritis, and duodenitis. Less common are the vascular malformations and iatrogenic factors due to endoscopic procedures and minimally invasive surgery.⁴

Hemobilia accounts for a small percentage of gastrointestinal bleeding. The first description in a patient with hepatobiliary trauma was made in 1948 by Sandblom.¹ There are few cases with atypical presentation as previously described. The risk of hemobilia after a cholecystectomy is less than 5% and is associated with endoscopic procedures.⁴ Vascular injury might provoke fistulas with the bile duct and cause acute gastrointestinal bleeding involving high-pressure vessels, which might compromise the hemodynamic status of patients.⁵ Approximately 50% of the cases are associated with invasive percutaneous procedures. According to the literature, 38% are linked to biopsies or percutaneous procedures; 19% to minimally invasive or open surgery, and only 10% to endoscopic intervention.⁶

The specific traumatic events for hemobilia can be injuries from diathermy, sutures or clips that might disrupt the vascular wall and the bile duct.⁷ Less common causes include inflammatory processes or infections.⁸ They are classified in Figure 5. Bile has fibrinolytic properties that might erode the mucosa or the vascular wall and delay healing. When combined with clots, it decreases their adhesion and might generate biliary obstruction or gastrointestinal bleeding, reported as melena or hematemesis.⁹

Clinical manifestations are conditioned by the magnitude of bleeding. Although there is a clinical trial, its presence is limited to 22% of reported cases.³ Bleeding typically presents as episodes of melena in 90% of cases and hematemesis in 60%. In the current patient, bleeding was evident through the Kerr tube. If hemobilia presents late, it might be associated with a traumatic event. It is difficult to achieve a diagnosis if there is no clear evidence of biliary pathology. However, the current patient had evident bleeding through her Kerr tube and clear clinical features. Therefore, the approach was focused on hemobilia. If there is no evidence for such etiology, it is necessary to perform an endoscopic evaluation to rule out different causes. It is possible to find bleeding or clots through the ampulla of Vater in 90% of cases.¹⁰

The endoscopic approach may be diagnostic in up to 60% of cases. Commonly reported features include the presence of fresh bleeding or clots, filling defects, and common bile duct or perihilar dilatation. The availability of endoscopic ultrasound allows for ruling out vascular etiologies due to portal hypertension.⁷ Imaging tests such as angiography are relevant for identifying vascular malformations, enabling selective embolization treatment. Therefore, it is rarely described as a first-line surgery and is usually reserved for difficult cases in which intravascular or percutaneous therapy is not possible. The success rate is greater than 90%, but mortality exceeds 10%. Surgical approaches may include hepatic artery ligation, pseudoaneurysm excision, liver lobectomy or segmentectomy.¹⁰

The use of ERCP is described as a diagnostic and therapeutic approach in cases of bleeding with a cholestatic pattern on the liver function panel.¹¹ This patient was managed with an endoscopic approach through ERCP with a favourable outcome.

CONCLUSION

Hemobilia is an uncommon cause of mid-gastrointestinal bleeding. However, it involves a high morbidity and mortality rate. Therefore, it is important to consider this etiology in cases of gastrointestinal bleeding when there is no evident cause, as seen in the previously reported case. Additionally, it is necessary to implement guidelines in order to determine the most suitable approach and to consider minimally invasive procedures.

Funding: No funding sources

Conflict of interest: None declared

Ethical approval: Not required

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Cite this article as: García EM, Chávez PDR, Ortiz MAG, Zariñana VHF, Ruiz JFM. Hemobilia, an uncommon case in a patient with severe acute cholangitis: case report. Int J Res Med Sci 2025;13:385-8.