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A Rare Case of Extended Retroperitoneal Biloma Due to Spontaneous Perforation of Common Bile Duct, Mimicking a Strangulated Right Inguinal Hernia: A Case Report and Literature Review

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Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
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Patient: Male, 89-year-old
Final Diagnosis: Bile duct injury • retroperitoneal biloma
Symptoms: Diffuse abdominal pain • fatigue • nausea • vomiting
Medication: —
Clinical Procedure: —
Specialty: Surgery

Objective: Rare disease


Background: Biloma is the collection of bile outside the biliary tree as a result of visceral perforation. The most common site of disruption is the gallbladder, whereas common bile duct lesions usually occur following medical procedures or trauma. Spontaneous perforation of the common bile duct has been previously reported in the literature. Retroperitoneal biloma secondary to spontaneous perforation of the common bile duct is an extremely rare pathological entity. The purpose of this report is to inform clinical doctors of this rare entity, which can have fatal consequences for the patient.

Case Report: We present the case of an 89-year-old man who was hospitalized with symptoms of vomiting, nausea, fatigue, and diffuse abdominal pain. The clinical examination and blood tests revealed peritonitis, a finding which was confirmed by the computed tomography of the abdomen as a retroperitoneal fluid collection, extending from the region posterior to the duodenum and head of the pancreas to the right inguinal fossa. As the patient's clinical status deteriorated, an urgent laparotomy was performed, revealing the presence of retroperitoneal biloma secondary to spontaneous perforation of the common bile duct. The operation was never completed as the patient died during the operation.

Conclusions: The diagnosis of this entity is difficult and is made during surgery. A large spectrum of treatment approaches has been used, but, regardless of the method, the goal is to halt the spreading abdominal contamination with bile and to treat the associated biliary pathology.


Keywords: Retroperitoneal Space • Spontaneous Perforation • Common Bile Duct • Surgical Procedure

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Background

Biloma is the collection of bile outside the biliary tree and can be either encapsulated or not. Gould and Patel first used the term in 1979 describing an abdominally entrenched bile collection [1]. The formation of biloma is the result of a lesion in the biliary tree, which can be either intrahepatic or extrahepatic [2]. As a consequence, biloma can be intrahepatic, peritoneal, or retroperitoneal [2], with the latter being the most uncommon [2].

The most common site of lesion is the gallbladder [3]. Injury to the common bile duct mostly occurs after surgical, endoscopic, or traumatic interventions, although spontaneous perforation has also been reported [4-25]. This condition is extremely rare in adults, occurring more often in children, due to congenital anomalies [4-6,11,12].

Satake, in 1985, was the first to describe a retroperitoneal collection of bile secondary to spontaneous perforation of the common bile duct [8]. To date, 7 cases have been published in the literature, reporting retroperitoneal biloma due to spontaneous perforation of the common bile duct [8,10,15-18,25].

The diagnosis of this pathological entity is difficult and the main diagnostic modalities for evaluation are abdominal ultrasound and computed tomography of the abdomen [1,2,4,9,25]. Despite these efforts, the diagnosis is usually made during surgery [1,2,4,9,25].

We present the case of an 89-year-old man with a retroperitoneal biloma with considerable extension; as a consequence, a mass was detectable in the right inguinal region, mimicking a strangulated right inguinal hernia. The patient was treated via urgent laparotomy, and spontaneous perforation of the common bile duct was identified.

Case Report

We present the case of an 89-year-old man who was admitted to the hospital with symptoms of vomiting, nausea, fatigue, and diffuse abdominal pain. The symptoms began 6 days before his admission. His vital signs on admission were: temperature=36.7°C, heart rate=113 bpm, respiratory rate=25 bpm, and blood pressure=113/70 mmHg, SpO2=97%. The patient was pale but with good nutritional status (body mass index=24 kg/m²).

The medical history of the patient included high blood pressure, dyslipidemia, and chronic obstructive pulmonary disease. His medications were amlodipine for regulation of high blood pressure, statin for treatment of dyslipidemia, and budesonide

Table 1. Blood test results on admission.

Blood test markers	On admission	Normal value range
WBC	47.150	3.800-10.500/mL
Hct	43.1	41-53.8%
Hb	14.6	13.4-17.4 g/dL
PLT	390	150-400 K/ μ L
CRP	44.25	<0.5 mg/dL
UR	57	15-50 mg/dL
CR	1.49	0.7-1.3 mg/dL
K ⁺	4.5	3.5-5.1 mmol/L
Na ⁺	140	136-145 mmol/L
TA- BIL	3.5	0.3-1.2 mg/dL
D -BIL	2.41	0-0.5 mg/dL
SGOT	34	0-35 U/L
SGPT	26	0-55 U/L
GGT	139	0-50 U/L
ALP	162	40-150 U/L

WBC – white blood cells; Hb – hemoglobin; Hct – hematocrit; PLT – platelets; CRP – C-reactive protein; UR – urea; Cr – creatinine; Na⁺ – sodium; K⁺ – potassium; TA-BIL – total bilirubin; D-BIL – direct bilirubin; SGOT – serum glutamic oxaloacetic transaminase; SGPT – serum glutamic pyruvic transaminase; GGT – gamma glutamyl transferase; ALP – alkaline phosphatase.

for management of chronic obstructive pulmonary disease. He had no history of previous surgical interventions and he did not consume alcohol or tobacco, and he had no history of abdominal or thoracic trauma.

On inspection, he was ill-looking and demonstrated signs of dehydration. The clinical examination revealed sensitivity and tenderness in all abdominal quadrants, with bowel sounds being absent. On digital rectal examination, there was no blood or palpable mass detectable. The clinical examination of cardiopulmonary and urogenital systems revealed no abnormal signs, and, apart from a feeling of fatigue, there were no other findings of physical and neurological examinations. Additionally, in the right inguinal fossa and scrotum there was a mass, giving the impression of a strangulated right inguinal hernia, but without elicited pain on palpation.

The blood tests revealed elevated inflammatory markers (WBC: 47.150/mL with normal values 3.800-10.500/mL and CRP 44.25 mg/dL with normal values <0.05 mg/dL), jaundice (TA-BIL: 3.5 mg/dL with normal values 0.3-1.2 mg/dL and D-BIL: 2.41 mg/dL with normal values between 0-0.5 mg/dL), as well



Figure 1. CT scan of the abdomen showing a retroperitoneal fluid collection. The arrow is demonstrating the fluid collection around the right kidney.

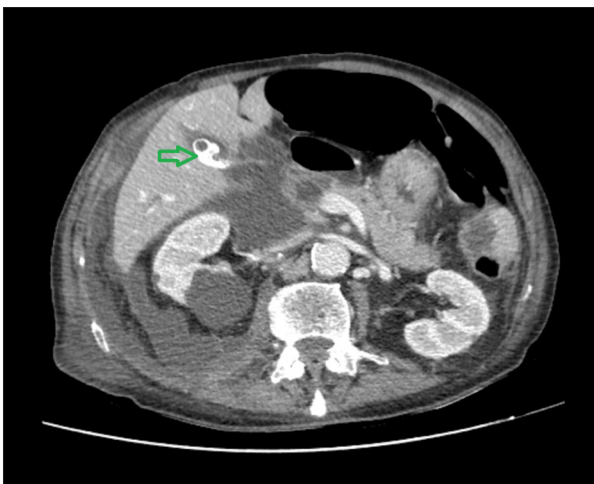


Figure 2. CT scan of the abdomen showing a retroperitoneal fluid collection, the gall bladder and the common bile duct. The arrow is demonstrating the gall bladder which is intact but with cholelithiasis.

as deterioration of the renal and liver functions (Ur: 57 mg/dL with normal values 15-50 mg/dL, Cr: 1.49 mg/dL with normal values 0.7-1.3 mg/dL, GGT: 139 U/L with normal values 0-50 U/L and ALP: 162 U/L with normal values 40-150 U/L). The results of all other laboratory markers were within normal range (**Table 1**). The blood cultures were negative for bacterial growth.

Taking into consideration all findings from the clinical examination and the blood tests, an abdominal X-Ray was performed, which was not diagnostic, followed by ultrasound of the abdomen and the right inguinal region. The former depicted cholelithiasis, without signs of inflammation or perforation of the gallbladder, and choledocholithiasis with normal diameter of the common bile duct (0.9 cm), while the latter revealed a fluid

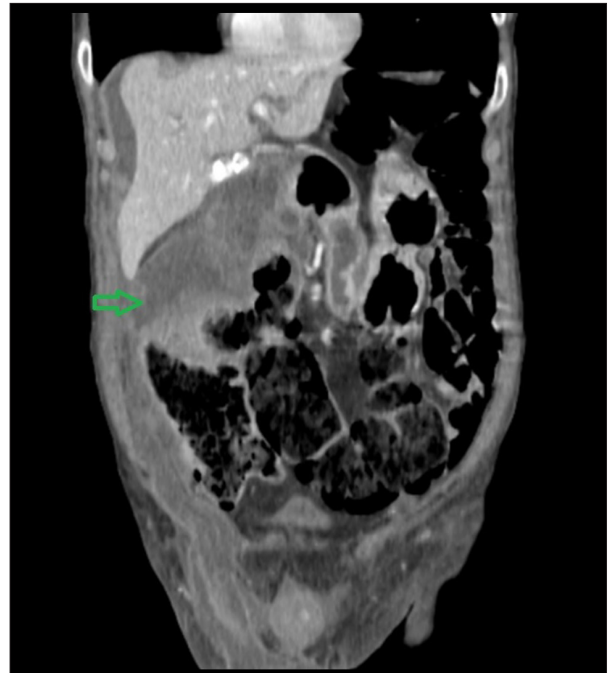


Figure 3. CT scan of the abdomen showing the extension of the retroperitoneal fluid collection. The arrow is demonstrating the collection which extends from the space dorsally to the duodenum and the head of the pancreas to the right inguinal region through Told's and Gerota's fascia, and right iliopsoas muscle.

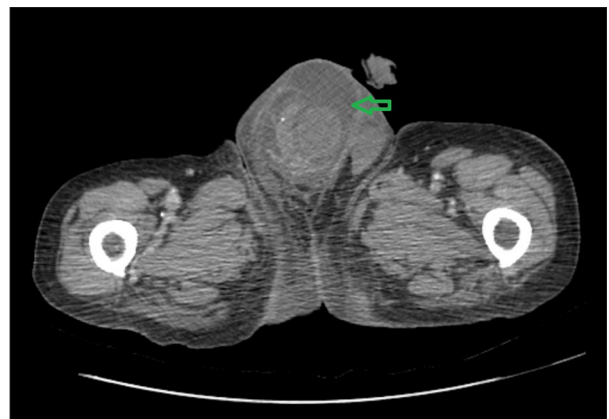


Figure 4. CT scan of the abdomen showing the extension of the retroperitoneal fluid collection in the right inguinal region and scrotum. The arrow is demonstrating the liquid which extends into the right inguinal canal where there were no omentum, mesenteric fat, or part of the bowel detected, and the scrotum.

collection in the right inguinal region and scrotum. The abdominal computed tomography confirmed these findings, additionally detecting a retroperitoneal fluid collection (**Figures 1, 2**). The collection was extending from the space dorsally to the duodenum and the head of the pancreas to the right inguinal

Table 2. Laboratory findings of the retroperitoneal fluid.

Biochemical analysis		Normal value range
TA-BIL	15.9	0.3-1.2 mg/dl
D-BIL	12.80	0-0.5 mg/dl
Microbial analysis		
Culture	Sterile	

region through Told's and Gerota's fascia, and right iliopsoas muscle (Figure 3). Furthermore, the liquid was extending into the right inguinal canal where there was no omentum, mesenteric fat, or part of the bowel detected (Figure 4).

Deterioration of the patient's clinical status with symptoms of vomiting, exacerbation of abdominal pain, and hemodynamic instability led to an urgent laparotomy with a midline incision 3 h after his admission to the hospital. The gallbladder was intact and there were no significant findings in the peritoneal cavity. A palpable mass was recognized at the right retroperitoneal space. The mobilization of ascending colon, hepatic angle and right mesocolon released substantial amount of green fluid, which was sent for biochemical and microbial analysis (Table 2). Through a Kocher's maneuver, the space located posteriorly to the duodenum and head of the pancreas was exposed and a small lesion of the common bile duct

was detected. The operation was never completed because the patient had cardiorespiratory arrest with subsequent unsuccessful resuscitation, and he died. Taking into account that the patient did not have any known coronary artery disease, we assume that sepsis was the main contributor to cardiorespiratory arrest.

Discussion

The most common site of perforation in the biliary tree, leading to the formation of a biloma, is the gallbladder [3]. The causes are typically cholelithiasis and cholecystitis [3]. McWilliams was the first to report 114 cases of perforation in the biliary tree in 1912 [21]. Most of these cases were in the gallbladder, 4 of them in the common bile duct, and 1 in the hepatic duct [21].

Injury to the common bile duct happens mostly after medical interventions or trauma, but a few cases of spontaneous perforation have been described [4-25]. It is a rare condition and the first to describe a non-traumatic perforation of the bile duct was Freeland in 1882 [13]. Chu reported 7 cases in 1984 and Kang another 11 in 2004 of spontaneous disruption of the common bile duct [4,14]. To our knowledge, these are the 2 largest series of non-traumatic perforation of the common bile duct ever to be reported.

Table 3. Comparison of the 7 cases with retroperitoneal biloma due to spontaneous perforation of the common bile duct, regarding the site of perforation and management. All 7 cases had cholelithiasis.

Author	Site of perforation	Management
Saravanan et al [18]	Not Found	<ul style="list-style-type: none"> • Percutaneous drainage • Endoscopic retrograde cholangiopancreatography
Yaşar et al [15]	Common bile duct	<ul style="list-style-type: none"> • Common bile duct exploration • T-tube intubation
Brady et al [16]	Not found	<ul style="list-style-type: none"> • Cholecystectomy • Common bile duct exploration • T-tube intubation
Satake et al [8]	Common bile duct	<ul style="list-style-type: none"> • Endoscopic retrograde cholangiopancreatography • Cholecystectomy • Common bile duct exploration • T-tube intubation
Hsieh et al [17]	Common bile duct	<ul style="list-style-type: none"> • Percutaneous drainage • Cholecystectomy • Common bile duct exploration • T-tube intubation
Blake-Siemsen et al [10]	Not found	<ul style="list-style-type: none"> • Endoscopic retrograde cholangiopancreatography • Common bile duct exploration • T-tube intubation
Takahashi et al [25]	Common bile duct	<ul style="list-style-type: none"> • Pancreatoduodenectomy

Several propositions have been made for the pathogenesis of this entity [3,9,10,14]. The most widespread is the increased intraductal pressure due to obstruction caused by gallstones, tumor, or reflux spasm of the sphincter of Oddi [3,9,10,14]. Other possible explanations include intramural infection of the common bile duct as a result of cholangitis, thrombosis of a mural vessel leading to necrosis of the affected part, diverticulitis of the common bile duct, acute pancreatitis, parasitic infections, and regurgitation of pancreatic secretions into the common bile duct [3,9,10,14].

If the site of perforation leads to the retroperitoneal space, a retroperitoneal bile collection will develop. A retroperitoneal biloma occurs as a result of bile tracking along the embryological fascial planes, with only 13 cases being reported [3,7,8,10,15-20,23-25]. To date, only 7 cases of a retroperitoneal biloma following spontaneous perforation of the common bile duct have been described in the literature [8,10,15-18,25] (Table 3).

In our case, the biloma was so extended that a mass in the right inguinal region, mimicking a strangulated right inguinal hernia was developed. This is an extremely rare condition. Only 1 similar case has ever been reported [16]. Brady in 2006 described a palpable mass in the right inguinal fossa in a 73-year-old man with a retroperitoneal biloma [16]. In some of these cases, the surgeons could not identify the exact site of perforation, despite an extensive examination [10,16,18]. The diagnosis of spontaneous perforation of the common bile duct was made during the surgical procedure from indirect evidence such as the site of biloma and the integrity of the gallbladder [10,16,18].

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In the management of a retroperitoneal biloma due to perforation of the common bile duct, various approaches have been described and a large spectrum of methods have been used [4,8-10,12,15-18,22,25]. The goals are to halt the spreading abdominal contamination with infected bile and to treat the associated biliary pathology [12]. The most frequent treatments to be reported are intraoperative drainage of the collection, ductal exploration, and T-tube intubation. An endoscopic approach using endoscopic retrograde cholangiopancreatography and percutaneous catheter drainage have also been used [4,8-10,12,15-18,22,25].

Conclusions

We report a rare case of retroperitoneal biloma following spontaneous perforation of the common bile duct. The diagnosis of this pathological entity is difficult and the management remains controversial. Having a high level of suspicion, performing appropriate imaging tests, and offering definitive treatment for this life-threatening condition as soon as possible is the only way to increase patient's chances for survival.

Department and Institution Where Work Was Done

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Declaration of Figures' Authenticity

All figures submitted have been created by the authors who confirm that the images are original with no duplication and have not been previously published in whole or in part.

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