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Spontaneous Retroperitoneal Biloma "Choleretroperitoneum" Complicating a Tumor of the Main Bile Duct: Exceptional Complication about a Case

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Abstract **Case Report**

Biloma refers to the accumulation of bile outside the biliary tract, which can be encapsulated or not. This condition is often caused by trauma, abdominal surgery, duodenal diverticula, or biliary disease. Spontaneous perforation of the common bile duct is rare in adults, and only a few cases have been reported in the literature. We present a case of spontaneous retroperitoneal biloma complicating a tumor of the main bile duct in a 77-year-old woman.

Keywords: Choleretroperitoneum - biloma - tumor of the main bile duct – emergency**.**

INTRODUCTION

The term "biloma" refers to the accumulation of bile fluid outside the biliary tract, which can be encapsulated or not. This condition is often caused by trauma, abdominal surgery, duodenal diverticula, or biliary disease. It rarely occurs spontaneously, and its rupture in the retroperitoneal cavity is exceptional (Kaushik & Attri, 2004). Spontaneous perforation of the main bile duct is quite common in the pediatric population, but it is very rare in adults, and only a few cases have been reported in the literature (Khanna *et al.*, 2010). In this article, we report an interesting case of spontaneous perforation of the main bile duct in an elderly woman and review the literature on similar cases.

OBSERVATION

This concerns a 77-year-old woman with no particular medical history. She presented to the emergency department with right upper quadrant pain and right iliac fossa pain, accompanied by vomiting and liquid diarrhea that had been ongoing for 15 days. All of this occurred in the context of low-grade fever and maintaining general health. Physical examination revealed sub-icterus, pain upon palpation of the right upper quadrant, and tenderness in the right iliac fossa. Her blood test results showed anemia at 10.9 g/dl, hyperleukocytos is at 21000, and a disturbed hemostasis with a prothrombin time of 45.2%. Her CRP was 125 mg/L, and lipase was normal.

Abdominal ultrasound showed a large heterogeneous collection visible from the right upper quadrant to the right iliac fossa, with diffuse infiltration of mesenteric fat. The gallbladder was multilithiasic, with dilation of the intra and extra hepatic bile ducts, including the common bile duct with a diameter of 17mm. The ultrasound also revealed two homogenous pseudotumoral cystic formations in the V segment with minimal peritoneal effusion.

Abdominopelvic CT scan revealed a large retroperitoneal collection on the right side extending from the hepatic hilum to the pelvic area. The collection was hypodense and not enhanced after contrast injection **(figure 1)**.

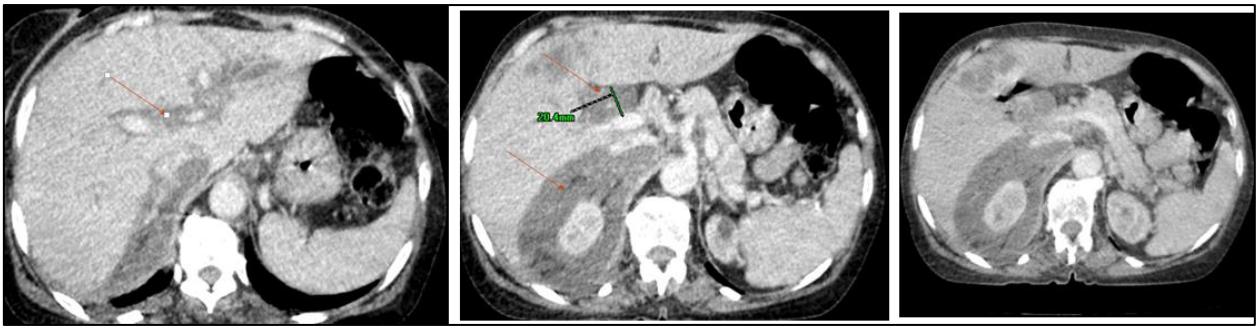

Figure 1: Abdominal CT scan with contrast injection: Dilatation of the main bile duct measuring 20mm, and of the intrahepatic bile ducts. The retroperitoneal collection (choleretroperitoneum) is spontaneously hypodense and not enhanced after contrast injection

The liver was of normal size and had four spontaneously hypodense cystic formations that were enhanced peripherally after contrast injection **(figure 2)**, with dilation of the intra and extra hepatic bile ducts **(figure 5-6)**. The appendix was swollen and measured 10mm with significant infiltration of mesenteric fat.

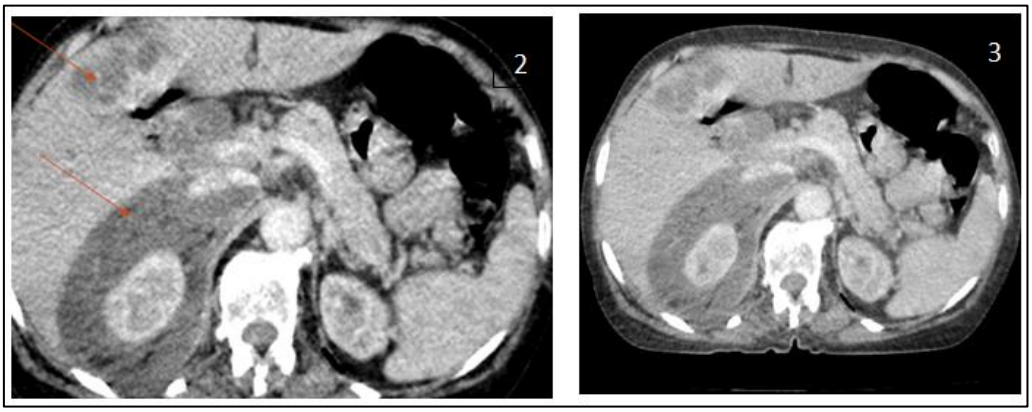

Figure 2-3: Abdominal CT scan with contrast injection: Dilatation of the intrahepatic bile ducts with individualization of two hepatic cystic formations (metastases on anatomical examination) enhanced in the periphery after contrast injection.

An echo-guided puncture of the collection retrieved a greenish cloudy fluid with a bile-like appearance **(figure7)**

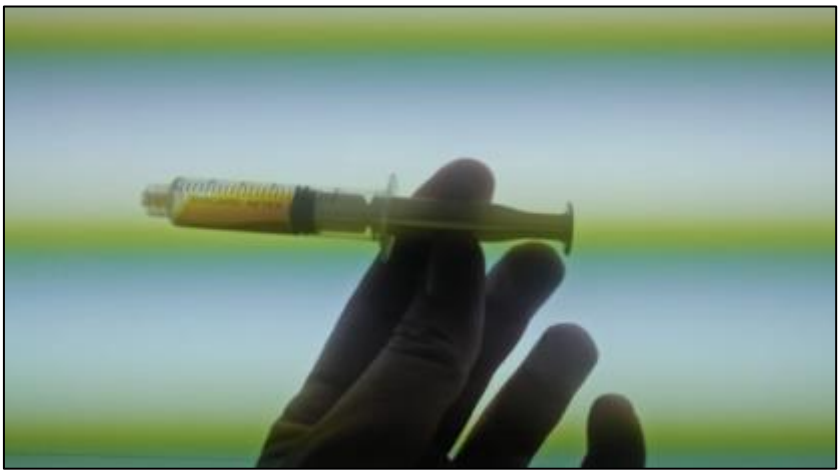

Figure 4: Echo-guided puncture of the collection brought back a greenish viscous fluid similar to bile.

The patient was urgently transported to the operating room for surgical exploration, which revealed a sclero-atrophic gallbladder in pyocholecystitis with a dilated common bile duct measuring 15mm. There was also a parietal defect responsible for a large retroperitoneal biloma on the right side. The surgical procedure involved cholecystectomy and restoration of the continuity of the bile ducts, as well as the removal

of the hepatic, epiploic, and peritoneal nodules, and aspiration of the retroperitoneal collection on the right side **(figure 5-6-7)**.

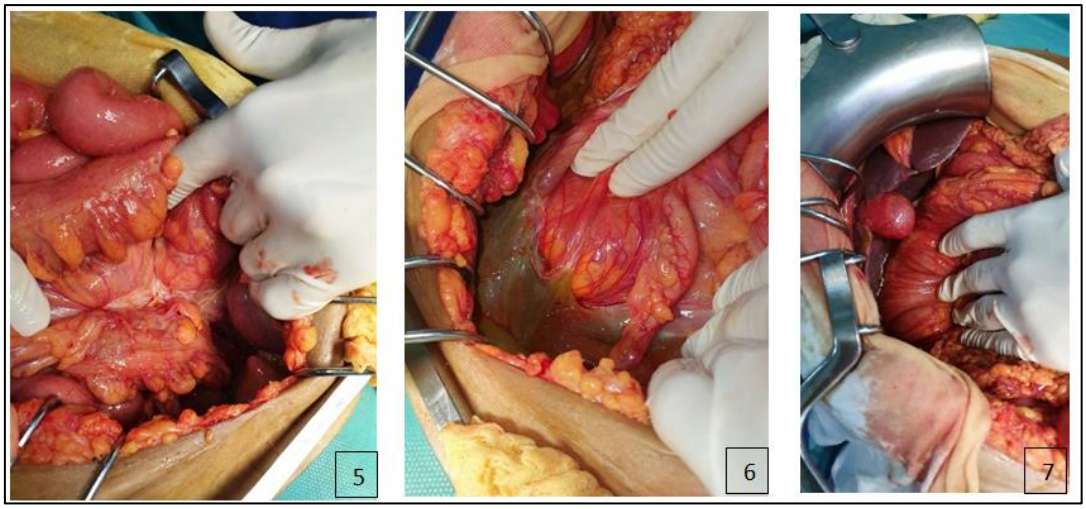

Figure 5-6-7: Images from the operating room showing the retroperitoneal collection with the liver metastases of segments IV and V.

The pathological results showed a poorly differentiated adenocarcinoma of the gallbladder that had infiltrated the entire wall of the hepatic parenchyma.

DISCUSSION

Spontaneous perforation of the extrahepatic bile ducts is extremely rare and very few cases have been reported in adults. In a literature review of spontaneous perforation of the bile ducts in adults, 70 cases were found. Among these, the site of perforation was the common bile duct in 42 cases, followed by the hepatic duct in 28 cases, frequently caused by a stone in the common bile duct followed by complications of bile duct surgery (Kang *et al.*, 2004a)

The retroperitoneal biloma, which means the accumulation of bile fluid in the retroperitoneal cavity, is an exceptional complication in adults (Kaushik & Attri, 2004). It is a rare entity, with few cases reported in the literature. The first case was described in 1882 by Freeman (*Freeman J. Rupture of the hepatic duct. Lancet. 1882;1:731---2.*, s. d.), and so far, the number of cases of choleretroperitoneum reported in the literature does not exceed 12 cases.(Blake-Siemsen & Kortright-Farias, 2017)

It is difficult to determine the responsible etiology because sometimes it is impossible to determine the site of the perforation. The retroperitoneal portion of the common bile duct remains the most likely source of the bile leak *(Kang et al.*, 2004b).

The clinical presentation can be acute or insidious, with the latter being more common and manifesting as abdominal pain associated with a progressively evolving jaundice and sometimes abdominal distension. In the acute form, patients are admitted in a state of cholangitis, cholecystitis, or biliary peritonitis (Haller *et al.*, 1989).

Generally, a bile leak into the intraperitoneal space presents as a picture of bile peritonitis, but in the case of a retroperitoneal accumulation, the peritoneal cavity may not be affected, as observed in our patient and in the case reported by Blake-Siemsen *et al.*, (Blake-Siemsen & Kortright-Farias, 2017).

Abdominal ultrasound is the first examination that can guide us towards the diagnosis of retroperitoneal biloma, it can show a perirenal, retroperitoneal, anechoic or heterogeneous collection in the event of superinfection. A dilation of the bile ducts is observed in most cases, as in the case of our patient. The differential diagnosis at this stage can be made with a retroperitoneal abscess or a chronic liquefied hematoma (Saravanan *et al.*, 2007).

Abdomino-pelvic computed tomography confirms the location of the collection, its extent, the integrity of the peritoneal organs, and the condition of the bile ducts. In the majority of cases, the CT scan helps diagnose and rule out intra-peritoneal involvement, but it does not confirm the etiology of the bile leak, as in the case of our patient.

Ultrasound-guided aspiration appears to be very useful because it confirms the nature of the fluid in the collection and suggests the biliary origin of the collection. In our case, as in the cases reported by Blake-Siemsen *et al.*, (Blake-Siemsen & Kortright-Farias, 2017) et Saravanan *et al.*, (Saravanan *et al.*, 2007), ultrasound-guided aspiration of the fluid played a major role in confirming the biliary origin and proposing the diagnosis of retroperitoneal biloma

"choleretropneum". The particularity of our case is that the cause of spontaneous perforation was caused by an adenocarcinoma of the main bile duct, with an insidious evolution of abdominal symptoms, whereas lithiasis is the responsible origin of perforation in the cases reported in the literature (Ishii *et al.*, 2016), (Kaushik & Attri, 2004), (Khanna *et al.*, 2010), (Blake-Siemsen & Kortright-Farias, 2017)

As in the case of our patient, in most cases, the diagnosis of cholérétropéritoneum is usually made during surgery, which allows for good visualization of the parietal defect and precise identification of the mechanism responsible for the perforation.

CONCLUSION

Choléretroperitoneum due to spontaneous perforation of the common bile duct is a rare and difficult to diagnose complication. It should be considered in the differential diagnoses of a retroperitoneal collection, and priority should be given to ultrasound-guided puncture of the fluid to provide a good diagnostic orientation and ensure prompt and adequate management.

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