

Spontaneous Biloma Secondary to Choledocholithiasis

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ABSTRACT

A biloma is a collection of bile located outside the bile duct which occurs as a result of iatrogenic and traumatic injuries. Spontaneous biloma is rare and is associated with choledocholithiasis. Diagnosis is performed using an ultrasound, a computed tomography scan, and a nuclear magnetic resonance scan, and is confirmed by drainage and subsequent biochemical analysis of the fluid sample. The first treatment option is percutaneous drainage, and if not successful, endoscopic biliary drainage should be performed. We report a case of a 46-year-old patient with a spontaneous biloma associated with choledocholithiasis.

INTRODUCTION

A biloma is defined as a collection of bile located outside the bile duct, and may be intrahepatic or extrahepatic. Bilomas are mainly caused by iatrogenic or traumatic injuries. However, spontaneous bilomas have also been reported. We report a case of a patient with spontaneous biloma associated with choledocholithiasis treated with percutaneous drainage and endoscopic biliary decompression.

CASE REPORT

A 46-year-old woman with a history of laparoscopic cholecystectomy in 2015 was referred to our department to evaluate for obstructive jaundice without signs and symptoms of cholangitis. An abdominal ultrasound revealed a dilated common bile duct with multiple stones without perihepatic fluid collection. After 48 hours, the patient developed sudden epigastric pain associated with a palpable abdominal mass. A computed tomography (CT) scan was performed showing a hypodense subcapsular image, perihepatic and subhepatic fluid spreading to the pelvis, and dilation of the common bile duct with choledocholithiasis, without evidence of other focal liver images (Figure 1). Because there was no definitive diagnosis, a CT-guided percutaneous drainage of fluid collection was performed, and 2,150 mL of bile was aspirated. The diagnosis of spontaneous biloma secondary to choledocholithiasis was made.

The patient showed clinical improvement. An endoscopic retrograde cholangiopancreatography (ERCP) was performed, revealing no dilation of the bile duct with multiple filling defects of the bile duct and no evidence of contrast material extravasation (Figure 2). A biliary sphincterotomy was made, and multiple stones larger than 10 mm were extracted with Dormia basket. A control cholangiogram showed no filling defect of the bile duct. After 3 weeks, the indwelling percutaneous catheter was removed, and the patient was discharged from the hospital.

DISCUSSION

A biloma is defined as a collection of bile located outside the bile duct. It can be intrahepatic or extrahepatic, and it may be encapsulated or not. Bilomas have a high mortality rate unless promptly diagnosed.¹ The incidence rate is 0.3%–2%, with no difference between sex.² This condition is found more often in the sixth and seventh decades of life.³ The most frequent location of bilomas is intrahepatic or in the right subphrenic or subhepatic spaces.¹ Bilomas are caused by iatrogenic or traumatic injuries. The most frequent cause is iatrogenic because of cholecystectomy, ERCP, liver biopsy, radiofrequency ablation, and transcatheter arterial chemoembolization.¹

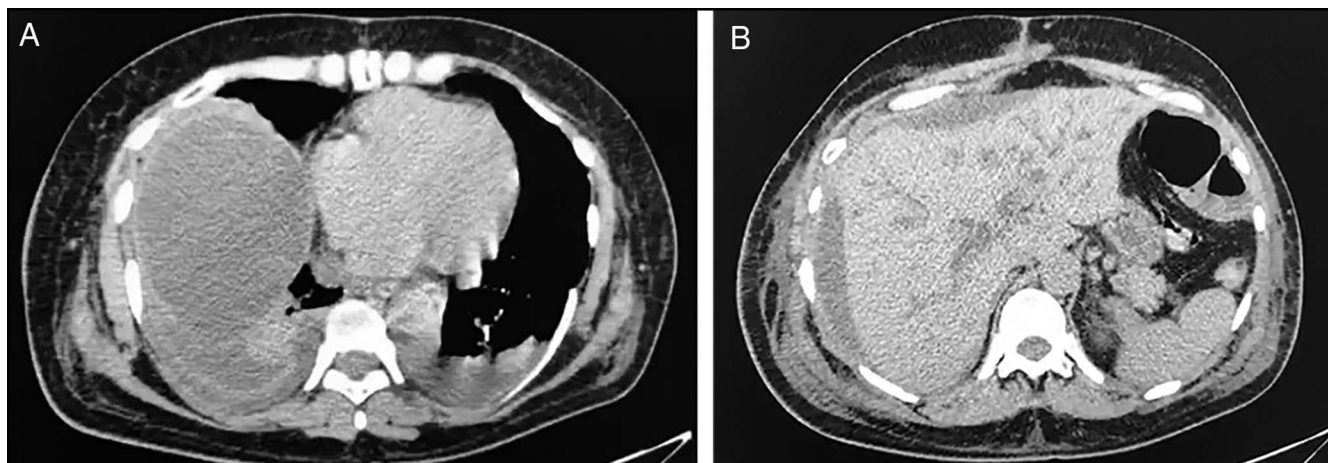


Figure 1. Computed tomography scan with intravenous contrast in the axial plane showing (A) hypodense subcapsular and (B) dilated intrahepatic bile duct with perihepatic fluid.

Rarely, bilomas resulting from biliary obstruction because of choledocholithiasis, cholangiocarcinoma, acute cholecystitis, hepatic abscess, tuberculosis, and sickle cell disease have been reported.⁴ In our case report, the spontaneous biloma developed during hospitalization as a result of an abrupt increase in bile duct pressure secondary to biliary obstruction because of choledocholithiasis. Iatrogenic lesions of the biliary system are more frequent when performing a laparoscopic cholecystectomy. After surgery, the average time to develop a biloma is approximately 2 weeks.⁵ In our case report, biloma was not associated with the cholecystectomy since there was a period of 5 years between both events. Radiofrequency ablation is associated with a 4% rate of biloma formation.^{1,6}

Choledocholithiasis is the most common cause associated with the formation of spontaneous biloma, arising from an increase in intraductal pressure because of stone obstruction and a weak bile

duct wall.⁷ Clinical presentation includes pain in the right upper quadrant, nausea, vomiting, and, in some cases, fever and jaundice if there is extrinsic compression of the bile duct. Some patients may present with septic shock, peritonitis, biliopleural fistula, bilhemia, hemobilia, and arterial pseudoaneurysm rupture.² Biochemical findings are nonspecific and include neutrophilic leukocytosis and cholestasis.¹

Abdominal ultrasound is often used as first-level imaging because patients tend to present with right upper quadrant abdominal discomfort.^{1,4} Findings suggesting a biloma include anechoic fluid collection with low-level internal echoes, well-defined margins, and absence of vascularization in Doppler ultrasound. The presence of internal septa suggests an infected biloma.¹ These findings are nonspecific; therefore, a differential diagnosis may include lymphocele, abscess, hematoma, hepatic cysts, cystic seromas, and cystic peritoneal metastases. In such cases, a CT scan with intravenous contrast and magnetic resonance (MR) scan are helpful for identifying, localization, and definition of their underlying cause. In a CT scan, bilomas usually reflect well-defined hypodense margins, with baseline values from 5 to 25 Hounsfield units, with no enhancement after the administration of contrast. In a MR scan, a biloma may present a heterogeneous signal intensity in a T1-weighted image and a homogeneous hyperintensity in a T2-weighted image.⁵ In some cases, an MR cholangiopancreatography might be helpful to locate the site of biliary leakage.¹

Although clinical history, location, and the imaging appearance of bilomas are sufficient at times, needle aspiration is often required to obtain a definitive diagnosis of bilomas as well as to direct clinical management.¹ Treatment of bilomas is not always necessary; a more conservative approach may be sufficient. Spontaneous reabsorption of bile collections larger than 4 cm is found to be rare and unpredictable.⁸ Most bilomas require treatment with a multidisciplinary approach, including surgeons, gastroenterologists, and interventional radiologists.⁹ Antibiotics in case of infection and drainage of bilomas are the



Figure 2. Endoscopic retrograde cholangiopancreatography revealing a thin bile duct with stones inside.

mainstay of treatment. The pathogenic spectrum includes *Enterobacteriaceae* and *Enterococcus* spp.¹⁰ Nowadays, treatment for spontaneous bilomas is mainly nonsurgical, involving percutaneous drainage with a pigtail catheter. Biliary decompression through ERCP is performed to ensure the resolution of the biloma through endoscopic sphincterotomy with or without stent placement.¹¹

Drainage through endoscopic ultrasonography is a new option, which is technically feasible and safe, and it provides an attractive alternative to percutaneous or surgical drainage.^{9,12} Surgery is reserved for patients in whom endoscopic stone extraction has failed or when there is a persistent active leak.¹¹ It has shown excellent outcomes. Nevertheless, it also has a high morbidity rate.⁹ In conclusion, a spontaneous biloma secondary to choledocholithiasis is an unusual disease. An early diagnosis is essential to prevent potentially life-threatening complications. Treatment involves a multidisciplinary approach.

DISCLOSURES

Authors contributions: M. Arramón and S. Martín wrote the manuscript. GJ Correa, M. Yantorno, A. Redondo, F. Baldoni, and F. Tufare approved the final version of the manuscript. M. Arramón is the article guarantor.

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