

## Spontaneous hepatic subcapsular biloma: Report of three cases with review of the literature

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*A biloma is an encapsulated bile collection outside the biliary tree. Most cases are caused mainly by iatrogenic injury and trauma, and are usually located in the sub-hepatic space. Spontaneous biloma is an uncommon entity. We report three rare cases of spontaneous hepatic subcapsular biloma formation in association with choledocholithiasis in two patients and cholangiocarcinoma in one patient. All the patients presented with extrahepatic biliary obstruction with no previous history of abdominal surgery, instrumentation, or trauma. Ultrasound and computerized tomography of the abdomen documented hepatic subcapsular biloma. The patients were initially managed with antibiotics and radiologically guided pigtail drainage of the collections, followed by definitive treatment of their underlying cause.*

**Key words:** Biloma, hepatic, subcapsular, choledocholithiasis, cholangiocarcinoma

### Spontan hepatik subkapsüler bilioma: Üç vakanın sunumu ve literatürün gözden geçirilmesi

*Bilioma, safra yolu dışında yerleşen kapsüllü safra koleksiyonudur. Bunların büyük çoğunluğu iatrojenik nedenlere veya travmaya bağlı olarak gelişir ve genellikle subhepatik boşlukta yerleşirler. Spontan bilioma nadir görülen bir durumdur. Burada nadir görülen, ikisi koledokolitiazise ve biri kolanjiokarsinoma sekonder gelişen spontan hepatik subkapsüler bilioma vakası sunulmuştur. Tüm hastalar ekstrahepatik biliyer obstrüksiyon ile başvurmuştur ve hiçbirinde abdominal cerrahi öyküsü, enstrümantasyon veya travma öyküsü yoktur. Hepatik subkapsüler biliomaların tanımlanmasında ultrasonografi ve bilgisayarlı tomografi kullanılmıştır. Hastalar ilk olarak antibiyotik ve radyolojik klavuzluğunda pig-tail katater drenajı ile tedavi edilmiş ve ertesinde alt yatan etkenin özgül tedavisi yapılmıştır.*

**Anahtar kelimeler:** Bilioma, hepatik, subkapsüler, koledokolitiazis, kolanjiyokarsinoma

#### INTRODUCTION

The term “biloma”, denoting an encapsulated extrahepatic collection of bile, was first introduced by Gould and Patel in 1979 (1). The main causative factors of biloma formation are traumatic or iatrogenic injuries, including abdominal surgery, percutaneous catheter drainage, transhepatic cholangiogram, and endoscopic retrograde cholangiopancreatography (ERCP), and infrequently without an apparent cause (2).

Non-traumatic bilomas are commonly referred to as spontaneous bilomas. The most common underlying cause of spontaneous biloma is choledocholithiasis (3). Other rare causes include bile duct tumors, hepatic infarction and idiopathic. The mechanism of spontaneous biloma formation is thought to be either the raised intraductal pressure secondary to extrahepatic biliary obstruction (EHBO), rupture of a cyst/diverticulum, or a focal

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liver infarction (3, 4). We report three cases of spontaneously formed hepatic subcapsular bilomas secondary to EHBO, and we discuss how to diagnose and manage such patients, together with a review of the related literature.

## CASE REPORTS

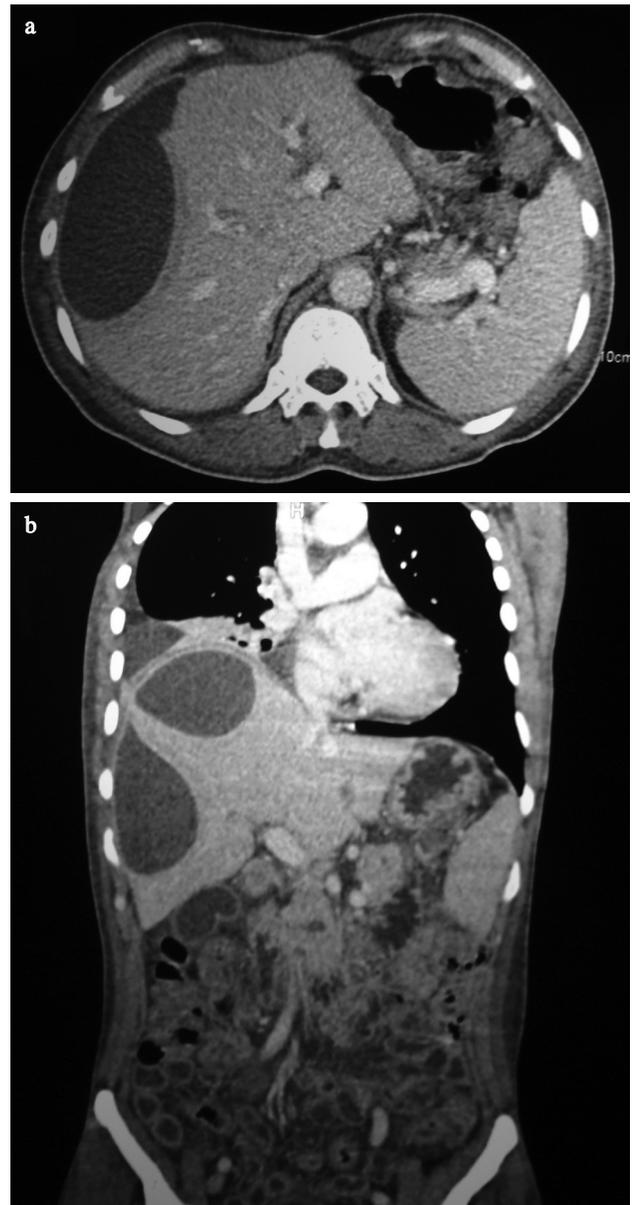
### Case 1

A 55-year-old man was admitted with intermittent colicky abdominal pain, fever and jaundice. He had no previous history of abdominal surgery, instrumentation or trauma. His general physical examination revealed icterus. On the abdominal examination, there was mild tenderness in the right upper abdomen with no palpable lump or organomegaly. Prophylactic broad spectrum antibiotics were started on admission. Laboratory investigations revealed: hemoglobin: 12.6 g/dl, white blood cells:  $12,500/\text{mm}^3$  (87% neutrophils), blood urea: 33 mg/dl, and creatinine: 1.2 mg/dl. Liver function test revealed a total bilirubin of 10 mg/dl with a conjugated component of 5.6 mg/dl; aspartate aminotransferase (AST) and alanine aminotransferase (ALT) were mildly raised. Alkaline phosphatase (ALP) was grossly increased (1060 mg/dl). The chest X-ray revealed right plural effusion and a raised right hemidiaphragm. Ultrasonography (USG) of the abdomen revealed a large unilocular subcapsular fluid collection measuring 9 x 8 x 5 cm, going around the right lobe of the liver anterolaterally and reaching the bare area. There was no free fluid in the abdominal cavity. The common bile duct (CBD) was 13 mm in diameter with a hyperechoic shadow at its lower part. Intrahepatic biliary radicals (IHBR) were also dilated. Thoracoabdominal computed tomography (CT) confirmed a large hepatic subcapsular biloma with volume of approximately 230 ml (Figure 1a, 1b) with a CT number of 18 Hounsfield units (HU). Based on the imaging findings, the patient was subjected to USG-guided pigtail catheter drainage of the collection, which drained about 160 ml of bile during the first 24 hours (h). Minimal drainage was observed over the next few days. Endoscopic sphincterotomy was done, and a 13 mm solitary stone was retrieved. The subcapsular fluid collection resolved, and the external percutaneous pigtail catheter was removed after five days. No residual or recollection was seen on follow-up USG at two and six weeks.

### Case 2

A 65-year-old woman was admitted with complaints of abdominal pain, nausea, intermittent vomit-

ing, fever, and yellowish discoloration of the eyes of seven days' duration. There was no significant medical history and no history of abdominal surgery, instrumentation or trauma. She was jaundiced and febrile. There were no respiratory or cardiovascular abnormalities. Abdominal examination revealed scratch marks on the abdominal wall, with mild diffuse tenderness. There were no other abnormalities on the examination. Initial investigations revealed a white cell count of  $13,500/\text{mm}^3$



**Figure 1a and 1b.** Contrast-enhanced computed tomography (CECT) of the abdomen showing a large unilocular subcapsular fluid collection around the right lobe of the liver anterolaterally and reaching up to the bare area with a volume of 230 ml. Common bile duct (CBD) and intrahepatic biliary radicals (IHBR) are dilated.

with neutrophilia (91%). Other blood counts were otherwise normal as were blood urea, serum creatinine and electrolytes. Liver function tests revealed: bilirubin: 9.5 mg/dl (normal: 0.3-1.0 mg/dl) with direct component of 5.5 mg/dl, ALP: 680 (normal: 20-90 IU/L), AST: 180 U/L (normal: 5-45 U/L), ALT: 290 U/L (normal: 5-40 U/L), and gamma-glutamyl transpeptidase (GGT): 87 U/L (normal: <65 U/L), with a normal albumin and coagulogram. Ultrasound examination done at a peripheral hospital had reported ascites. USG of the abdomen was repeated in our hospital, which revealed a huge hepatic subcapsular collection extending all around the liver and causing severe compression of the liver parenchyma and IHBR. A large 18 mm hyperechoic shadow was seen in the distal CBD. Contrast-enhanced CT (CECT) of the abdomen confirmed a massive hepatic subcapsular collection of approximately 1500 ml (Figure 2a, 2b) with a CT number of 24 HU. CT-guided pigtail catheter drainage of the collection was done, which removed about 1200 ml of altered bile in the first 2 h after the procedure. The patient was relieved of acute pain immediately. The bile culture revealed a mixed growth of coli forms. The patient was already on broad spectrum antibiotics. Minimal drainage was noted after the first 48 h. ERCP revealed a dilated CBD and two stones (largest 20 mm) visible in the lumen. CBD exploration with choledocholithotomy and T-tube drainage was done, as ERCP and sphincterotomy failed to manage the choledocholithiasis. The pigtail catheter was removed during surgery. The postoperative period was uneventful, and the T-tube was removed on

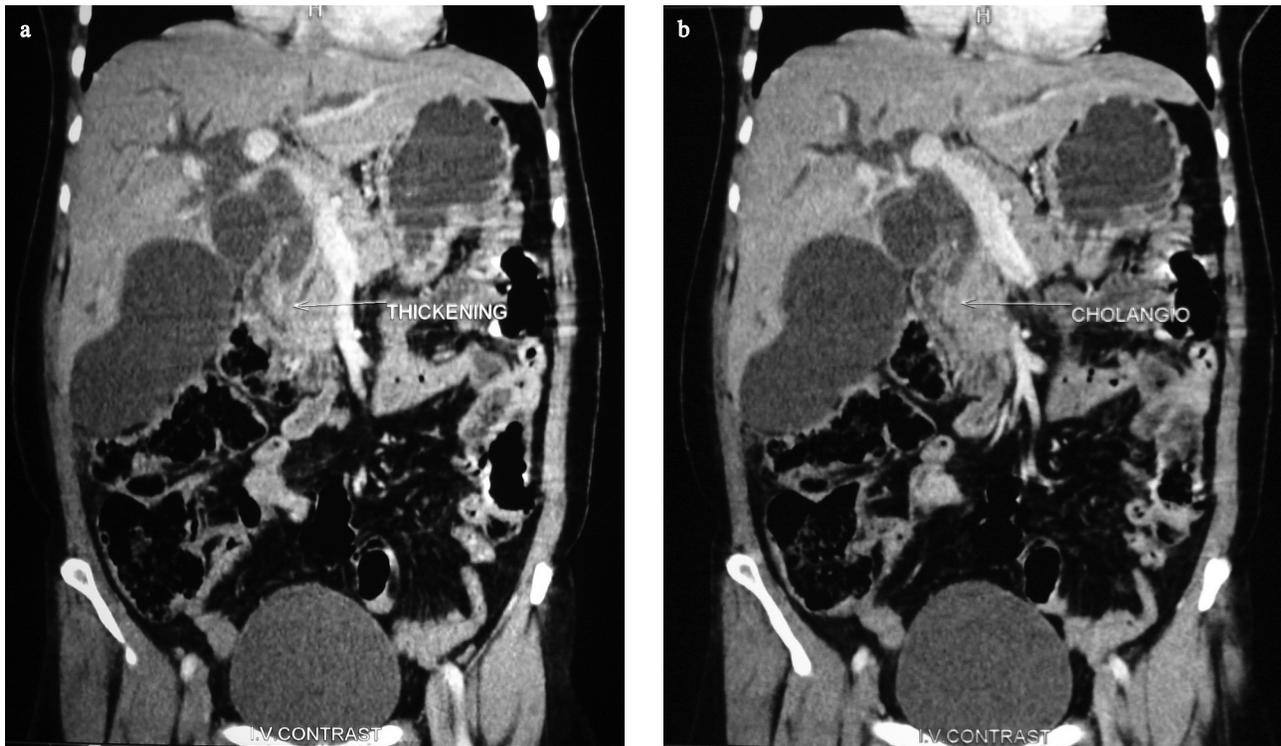
the 10<sup>th</sup> day after a normal T-tube cholangiogram. On follow-up, USG of the abdomen did not show any residual or recollection of bile.

### Case 3

A 50-year-old female presented with right upper abdominal pain and progressive jaundice of three weeks' duration. There was also a history of nausea, fatigue, anorexia, and weight loss. The patient had no history of abdominal surgery, instrumentation or trauma. On general physical examination, she had mild pallor and icterus. Abdominal examination revealed mild tenderness in the right hypochondrium with a palpable gallbladder. Cardiovascular and respiratory examinations were inconclusive. Complete blood count revealed only anemia (hemoglobin 8 g/dl). Liver function test revealed a total bilirubin of 12 mg/dl with a conjugated component of 7 mg/dl; AST and ALT were mildly raised, while ALP and GGT were grossly increased. Coagulogram was mildly deranged with international normalized ratio (INR) of 1.4. Other laboratory parameters were normal. USG of the abdomen revealed a subcapsular hepatic collection with dilated CBD and IHBR. On CECT of the abdomen, diffuse enhancing thickening of the lower CBD was noticed with abrupt cut-off at its lower end, along with a large hepatic subcapsular collection predominantly posteromedially along segment V1 of the liver. It was about 11 x 10 x 5 cm in size with a volume of approximately 300 ml and a CT number of 16 HU (Figure 3a, 3b). Broad spectrum antibiotics were started, and CT-guided pigtail catheter drainage of the collection was done, which removed



**Figure 2a and 2b.** CECT of the abdomen revealed a huge hepatic subcapsular collection (1500 ml) extending all around the liver and causing severe compression of the liver parenchyma and IHBR.



**Figure 3a and 3b.** CECT of the abdomen showing diffuse enhancing thickening of the lower CBD with abrupt cut-off at its lower end, along with a large hepatic subcapsular collection predominantly posteromedially along segment V1 of the liver (11 x 10 x 5 cm in size, with a volume of approximately 300 ml).

about 200 ml of bile over 24 h. Biliary decompression was achieved by endoscopic procedure with internal stenting of the CBD. The pigtail catheter was removed on the 6<sup>th</sup> day when there had been no drainage for 72 h. The patient was planned and prepared for surgery. At laparotomy, the tumor was found involving the lower third of the CBD with invasion of the periductal tissue, but without nodal involvement or distal metastasis (T2 N0 MO). Pylorus-preserving pancreaticoduodenectomy was done. Reconstruction was achieved by end-to-end pancreaticojejunostomy, end-to-side hepaticojejunostomy and end-to-side gastrojejunostomy. The postoperative period was complicated by left basal pneumonia, pulmonary embolism, hypoalbuminemia, and urinary infection. The patient's recovery was complete, and she was discharged on the 28<sup>th</sup> day and is presently under follow-up.

## DISCUSSION

The term biloma was introduced in 1979 by Gould and Patel to describe a loculated collection located outside the biliary tree. Kuligowska et al. (5) extended the term biloma to include intrahepatic as well as extrahepatic collections of bile.

Bilomas are most common after surgery, especially following cholecystectomy (laparoscopic/open), instrumentations including percutaneous transhepatic cholangiography, liver biopsy, biliary drainage procedures, ERCP, and trauma (2,4-7). Spontaneous formation of biloma is now a recognized clinical entity. The most common underlying cause of spontaneous biloma is choledocholithiasis. Other rare causes include bile duct tumors, hepatic infarction, abscess, and idiopathic (4).

Most of the collections are subphrenic and subhepatic. Subcapsular bilomas are less common and have been reported following surgery and trauma (2,4,8). Spontaneous hepatic subcapsular bilomas are seen very rarely.

The exact mechanism of spontaneous biloma formation is unclear. The suggested contributing factors are raised intraductal pressure caused by obstruction due to stone, tumor, and spasm of the sphincter of Oddi; necrosis of a part of the bile duct wall secondary to stone; rupture of a cyst or diverticulum; and focal liver infarction (3,4). In our patients, EHBO with raised intraductal pressure was present due to choledocholithiasis in two cases and cholangiocarcinoma in one case. The

subcapsular location of bilomas in these patients may be explained by the high intraductal pressures secondary to EHBO, associated with poor parenchymal support for distal biliary radicals, which open, causing leakage of bile contained by the hepatic capsule.

The usual presentation of biloma is right upper quadrant (RUQ) pain and abdominal fullness, which is at times associated with fever. Bilomas are more common in the RUQ of the abdomen, but can occur in the left upper quadrant in about 40% of cases, as the bile migrates from the RUQ to the left subhepatic or subphrenic space over the anterior part of the liver (3,4). The RUQ abdominal pain is the constant sign in the patient with subcapsular biloma described in the literature, associated sometimes with nausea and vomiting (8). Our patients in addition had clinical features suggestive of EHBO.

Ultrasound is sensitive for diagnosing bilomas, but the diagnosis of this complication is ideally facilitated by the use of CT (8). CT scan is optimal for identifying and localizing bilomas and showing their size, nature (unilocular or septate), distribution, and regional anatomy, as well as defining their underlying cause. The differential diagnosis includes hematoma, seroma, liver abscess, pseudocyst, liver cyst, and lymphocele. Anatomy of the collection and the CT number help in the diagnosis of biloma. Most bilomas have a CT number of less than 20 HU unless they are mixed with blood or exudates (9). USG and abdominal CT are sometimes unable to differentiate bilomas from seromas, lymphoceles and angiomas. Magnetic resonance imaging (MRI) or hepatobiliary scintiscan can be useful when continuous bile leakage is present, but they are not diagnostic when continuous leakage is not present. Radiological image-guided aspiration tests, when testing for bilirubin, can be diagnostic. When the aspirated substance is a clear yellow liquid, then microbiological tests must be done to rule out an infection. ERCP can be used to determine the location and severity of an active bile leak. However, the presence of small biliary

cysts or bilomas, located in the lower areas of the liver where they can be hidden by gastrointestinal shadows, can be difficult to diagnosis (10). USG and CT scan can also guide accurate drainage of the collections, as was done in all three patients reported herein.

Treatment for bilomas with a diameter of only a few centimeters is not always necessary; these lesions can be watched. However, most bilomas require treatment. In the past, surgery was the main approach to treatment (10). Currently, the treatment for spontaneous bilomas is mainly nonoperative, in the form of radiological percutaneous drainage with pigtail catheters, endoscopic nasobiliary drainage (ENBD) and endoscopic sphincterotomy or stenting to lower the pressure of the biliary channels and achieve removal of stone, if indicated. Surgery is only required for very few patients - in those in whom endoscopic retrieval of CBD stones fails, in those who require definitive treatment for biliary tumors, or when there is persistent active bile leakage in spite of percutaneous drainage (10, 11). Two of our patients had choledocholithiasis, of which one also required choledocholithotomy for his impacted stone, while the other was managed with pigtail catheter drainage of subcapsular collections and endoscopic stone retrieval. The patient with cholangiocarcinoma was managed initially with pigtail drainage of the bile collection and biliary decompression by stenting of the CBD, which was followed by a definitive surgery.

In conclusion, the biloma is a rare complication of traumatic and obstructive hepatobiliary diseases. Despite choledocholithiasis being the most common cause of EHBO, we have described two cases with an unusual complication of this condition apart from a case of cholangiocarcinoma causing biliary obstruction and spontaneous hepatic subcapsular collection. The patients should be managed initially with antibiotics and radiologically guided drainage of the bilomas. Surgery is now performed only in cases with a persistent bile leak or for treatment of an underlying disease (10).

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