

Case report

Traumatic Intrahepatic Biloma – A Rare Entity Mimicking Neoplasm of the Liver: A Case Report

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SUMMARY

Bilomas represent large collections of bile, produced due to a rupture of bile ducts. Most frequently, they are located in the perihepatic tissues, rarely intrahepatic ones. Usually, bile ducts rupture is caused by trauma, spontaneous or iatrogenic, or some underlying disease. The majority of bilomas cases are treated by means of drainage procedures. In spite of advanced diagnostic techniques, sometimes they can be misinterpreted as neoplasm. We report a case of biloma that was surgically removed due to suspicion of malignancy, which was histologically examined.

Key words: biloma, trauma, liver, histopathologic confirmation

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INTRODUCTION

Bilomas represent large collections of bile, usually located outside the biliary tree. Most frequently bilomas are located in the perihepatic tissues and structures adjacent to the liver, near the gallbladder or large bile ducts, although they can be rarely found inside the liver. They are formed as a consequence of bile duct wall destruction by rupture or necrosis, with bile spillage into the surrounding peribiliary tissues (1).

Damage of the bile duct can be caused by trauma, iatrogenically (usually during laparoscopy procedures) or during ischemia and infections of the biliary tree. Sometimes, it can also occur spontaneously (2).

Histopathologically, biloma consists of bile fluids, sometimes with necrotic detritus, admixed with or surrounded by inflammatory cells and histiocytes which are often transformed into xanthomatous cells. At the periphery of these collections, usually there is a fibrous tissue proliferation, making them look encapsulated (1).

Depending of their location and size, bilomas are usually treated with drainage or surgically. Cases of bilomas with histopathological confirmation are very rare. Small in size, intrahepatic bilomas can easily be misdiagnosed as primary or metastatic liver tumors and therefore need to be surgically removed.

We represent a case of intrahepatic traumatic biloma mimicking a primary or metastatic liver tumor.

CASE REPORT

A 34-year-old man was admitted to hospital for several days lasting nausea, chronic right hypogastric pain and occasional episodes of vertigo, followed by short syncopes. The patient did not report any substances or unusual food intake, or recent travels. He reported falling into a bathtub during a syncopal episode three weeks before being admitted to hospital, with contusions of the right side of the thorax and abdomen. The physical examination revealed the presence of deep palpation pain in the right hypogastrium and epigastrium. Laboratory tests parameters were within the normal range.

An ultrasound examination of the abdomen revealed multiple calculi inside the gallbladder, with the largest diameter of 7 mm, and also a heterogenous, hypo-anechoic rounded lesion in the VI hepatic segment, with a maximum size of 37 mm, with irregular contours. This focal lesion was confirmed by abdominal CT which showed hypodense lesion with significant enhancing after intravenous application of contrast agent, along with

the segmental dilatation of bile ducts in the V and VI hepatic segment and slight capsule retraction. The lesion's appearance indicated metastasis or a primary bile duct tumor. Other CT findings were within normal range, as well as serum tumor markers (CEA and Ca 19-9). The patient underwent liver segmentectomy, and a part of the VI liver segment was removed along with the lesion.

Postoperative examination confirmed nodular subcapsular mass in the resected liver specimen, 15x14x12 mm in size, yellowish with thin fibrous capsule, with a softened central mass (Figure 1.)

The lesion was serially sampled and the specimens were stained with standard hematoxylin-eosin and Trichrom-Masson techniques.



Figure 1. Subcapsular nodular yellowish mass, 15x14x12mm, with fibrous pseudocapsule

On histopathological examination, the lesion consisted of necrotic and bile stained central detritus mixed with and surrounded by an admixture of inflammatory cells, predominantly lymphocytes, histiocytes (many of them with bile-stained cytoplasm), xanthomatous cells and scattered „foreign body type“ giant cells (Figure 2, 3.). The admixture of necrotic tissue and inflammatory cells was surrounded with granulation tissue and proliferated fibrous tissue with thick collagen fibers, along with parts of segmental bile ducts with dilated lumina and reactive epithelium (Figure 4). The lesion was diagnosed as biloma, probably of traumatic etiology.

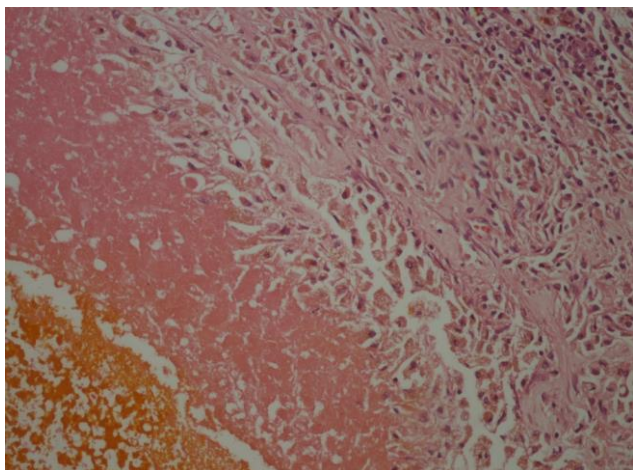


Figure 2. Bile stained necrotic detritus, surrounded by inflammatory cells, xanthoma cells and fibroblasts

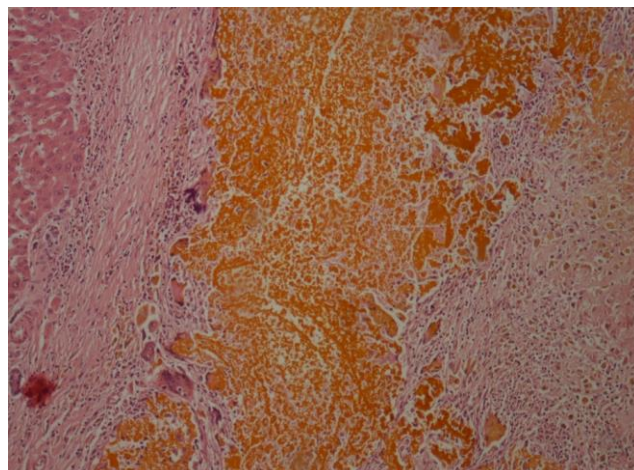


Figure 3. Foreign body-type giant cells surrounding necrosis and bile

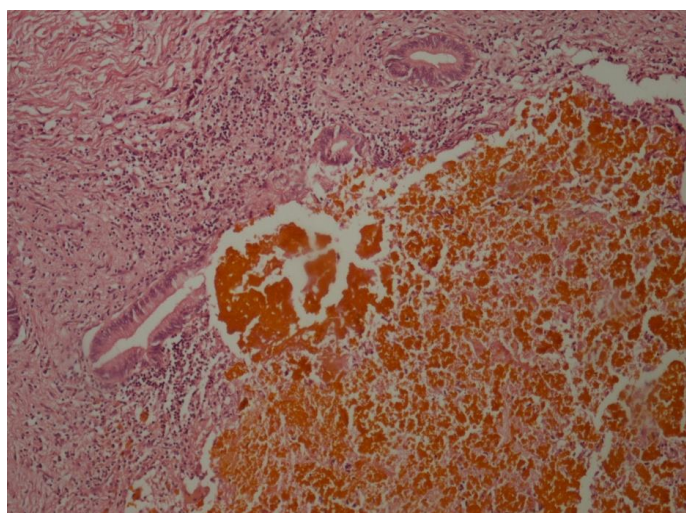


Figure 4. Remnants of ruptured bile ducts, surrounded by ductular reaction and inflamed fibrous tissue

DISCUSSION

The first case of biloma was described in 1979 by Gould et al. (3) as an encapsulated extrahepatic collection of bile. First post-traumatic case of biloma was reported by Whipple (4) who described a case of a man kicked by a horse. Gould reported a case with extrahepatic bile leakage after an abdominal trauma; the bile did not cause peritonitis, but it accumulated in the perihepatic tissue in an encapsulated form. This mechanism was later applied to all cases of biliary tract injuries with bile leakage that formed a capsule inside or outside the liver (3). The

encapsulation of bile is explained by a large amount of fast-leaking bile that causes biliary peritonitis, or a small amount of bile leaking slowly that causes mild inflammatory reaction. The leaking bile is simultaneously trapped by the mesentery and the greater omentum, where it becomes encapsulated. Even though the amount of bile leaked can be minor, the development of an inflammatory reaction contributes to the appearance of a growth in size of biloma (5).

In extremely rare cases bilomas develop spontaneously (6). They are mostly developed after some trauma (abdominal injury) or as an iatrogenic injury, du-

ring surgical or diagnostic procedures, such as cholecystectomy or endoscopic retrograde cholangiopancreatography. Sometimes, bilomas occur as a consequence of an underlying disease affecting the bile ducts: choledocholithiasis, cholangiocarcinoma, acute cholecystitis, liver infarcts (7-10, 11).

The size and location of bilomas depend on the main cause of bile duct rupture, on the speed of bile leakage and the speed of peritoneal resorption of bile. Various studies have shown that extrahepatic bilomas most often occur due to a rupture of the left hepatic duct and cystic duct; sometimes, the exact site of a bile duct rupture remains unknown. Intrahepatic bilomas are most commonly formed in the left hepatic lobe (5, 6).

Clinical symptoms of bilomas are often nonspecific. They can vary from the absence of symptoms to abdominal distention and pain, fever, jaundice and leukocytosis (12).

Diagnosis of biloma can be easily established with the presence of data about previous surgical procedure or trauma, and with an adequate ultrasound, CT or MRI examination. On abdominal ultrasound, bilomas are usually presented as hypoechoic masses with sharp borders, located around or inside the liver, eventually increasing in size with time (13). Abdominal CT provides more precise data about location of bilomas, as well as of their relations with adjacent structures and organs. Sometimes ultrasonography and CT are not confident for differentiating biloma from seroma, lymphocele or angiomatous proliferations. MRI or ERCP can be useful in cases with continuous bile leakage, but they are not particularly diagnostically specific if bile leakage is not present (9, 14). In some cases diagnosis can be established with aspiration biopsy of the lesion, with the presence of bilirubin in the aspirated liquid (15). ERCP can be used for detecting the exact site and intensity of active bile leakage.

With all available diagnostic procedures, bilomas are sometimes difficult to diagnose, especially if they are small in size (up to few centimeters), without active bile leakage or located in the lower portions of the liver (16).

Intrahepatic bilomas are rarely seen because of lesser risk of injuring intrahepatic bile ducts. It can occur after blatant traumas or penetrating abdominal injuries, as well as after intrahepatic surgery or therapeutic procedures (abscess drainage, RFA) (17-20).

Histopathological examination of bilomas usually requires staining of specimens with standard haematoxylin-eosin technique, which demonstrates characteristic yellowish-green bile aggregates. Additional histochemical or immunohistochemical techniques are rarely necessary (Trichrom Masson or Reticulin staining demonstrates fibrous tissues; Ki-67, CD68, or CD3 can be used for analyzing inflammatory cells).

For small bilomas (up to few centimeters in size), sometimes, therapy is not indicated. Some of them resolves spontaneously or after non-surgical procedures (ERCP with sphincterectomy or stent) (20-22). For larger bilomas, drainage procedures are the most frequent therapeutic option. Very few bilomas require surgical removal, usually in cases with continuous bile leakage or an underlying disease (21-23).

CONCLUSION

Traumatic intrahepatic bilomas occur rarely. In spite of advanced diagnostic procedures they can be easily misdiagnosed as other pathological processes, especially if they are small in size. Cases of histopathologically confirmed bilomas are extremely rare.

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Traumatski intrahepatični bilom- redak entitet koji može simulirati neoplazmu jetre: prikaz slučaja

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SAŽETAK

Bilomi predstavljaju kolekcije žuči nastale izlivanjem žuči usled ozlede bilijarnih vodova. Obično su lokalizovane u perihepatičnim strukturama, a ređe intrahepatično. Uzrok može biti trauma, jatrogeno oštećenje zida bilijarnog voda ili patološki proces. Uobičajena terapija je drenaža, ređe hirurško odstranjenje, a kod asimptomatskih malih biloma kliničko praćenje. U veoma malom broju slučajeva neophodna je patohistološka verifikacija. U ovom radu prikazan je slučaj intrahepatičnog biloma manjih dimenzija, nastalog usled traume, koji je operativno odstranjen usled sumnje na neoplastični proces u jetri.

Ključne reči: bilom, trauma, jetra, patohistologija