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Case Report

Spontaneous Subcapsular Biloma Presenting As Acute Abdomen: Case Report and Literature Review

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Abstract: A biloma by definition is an encapsulated collection of bile outside the biliary tree. It is mainly caused by iatrogenic injury or abdominal trauma. A spontaneous infected biloma without any underlying disease is a very rare finding. We report here a case of infected spontaneous biloma confirmed by ultrasonography, computed tomography and sonographically guided percutaneous aspiration and review the relevant literature.
Keywords: Biloma, Ultrasonography, Computed tomography.

INTRODUCTION

The term 'biloma' was first coined by Gould and Patel in 1979 [1]. It is the collection of bile outside the biliary tree. It can be seen at intra or extra hepatic location. It may or may not be encapsulated. It is most commonly iatrogenic, secondary to surgery or percutaneous transhepatic procedures like percutaneous transhepatic cholangiography (PTC),percutaneous transhepatic biliary drainage(PTBD) or may be secondary to abdominal trauma.

Spontaneous bilomas are very rarely described in the literature. This report present a case of spontaneous infected subcapsular biloma in middle aged man, who presented to us with acute abdomen and tachycardia.

CASE REPORT

A 51 years old male presented as acute abdomen in the emergency department of our institute with a pulse rate of 160/minute. He was having pain since past 2 days. He complained few episodes of vomiting. Past medical and surgical history was not remarkable.

At the time of examination he was afebrile, hemodynamically stable but he looked unwell. Abdominal examination revealed tenderness in epigastric region and right upper quadrant.

Initial investigations revealed mild leukocytosis (14700/mm^3). Liver function tests, amylase and coagulation screen were normal. An erect abdomen and chest radiographs were unremarkable. Ultrasound examination revealed hypo-anaechoic, heterogeneous collection at subcapsular location medial and inferior to left lobe of liver having multiple internal septae (Fig. 1). Other findings included cholelithiasis, mild (interbowel) peritoneal free fluid and bilateral mild pleural collection. There was no choledocholithiasis and common bile duct was normal in calibre. Computed tomography scan showed a well circumscribed collection of 8 x 6.6 cm size at subcapsular location medial, inferior and posterior to left lobe of liver (Fig. 2A) with extension of exudative inflammatory process in gastro-hepatic and hepato-duodenal ligament and mild (interbowel) peritoneal collection. On post contrast scan there was no enhancement (Fig. 2B). Additionally there was cholelithiasis, bilateral mild pleural collection and right inguinal hernia. On basis of imaging findings with clinico-pathological correlation provisional diagnosis of subcapsular biloma was made with differentials of abscess, biliary hydatid and seroma.

Ultrasound guided diagnostic aspiration from collection revealed the presence of a thick, dirty greenish material suggestive of infected bile. Biochemistry confirmed presence of bile within the fluid. Routine & microscopy revealed cell count of 1400/mm^3,of which,90% were neutrophils. Gram staining showed a gram negative bacillus. Negative serology for echinococus ruled out possibility of preexisting hydatid cyst of liver. After drainage of biloma, patient was kept on intravenous antibiotics and

he progressed well and was discharged on 7th day. Follow-up ultrasonography after 12 days revealed significant reduction in amount of collection. The patient was doing well clinically on follow-up visit after one month.

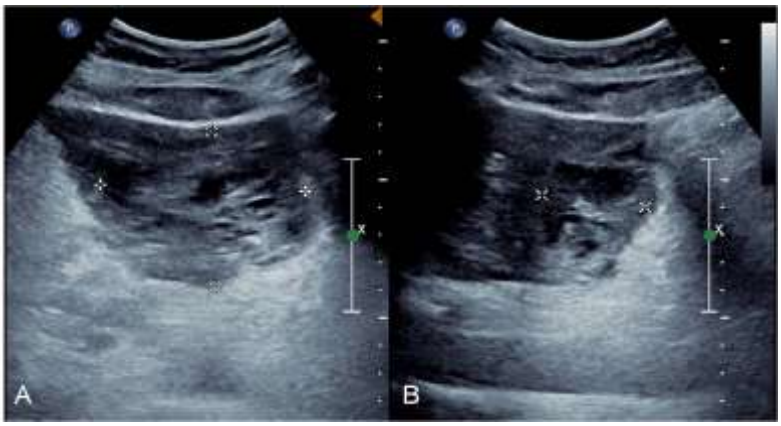

Fig. 1(A & B): Axial (A) and Oblique (B) ultrasound images revealing hypo-anechoic fluid collection with multiple internal septations, medial and inferior to left lobe of liver

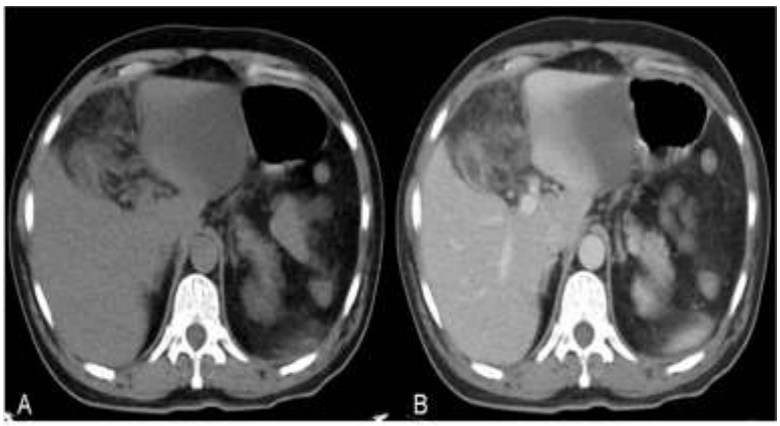

Fig. 2 (A & B): Axial C. T. Images, (A) Plain and (B) Post contrast revealing a fluid collection with absence of enhancement, medial and inferior to left lobe of liver

DISCUSSION

After the introduction of term 'Biloma' by Gould and Patel [1] in 1979, to describe an encapsulated bile collection located outside the biliary tree. The term was extended to include intrahepatic as well as extrahepatic collections of bile by Kuligowska *et al*. [2]. Bilomas are a rare entity and are most common after surgery/ interventional procedures/ trauma related to hepatobilliary system [3-5]. Spontaneous biloma is now a known entity in the literature, however there are only a few case reports. The exact mechanism of spontaneous biloma formation remains unclear [6]. Choledocholithiasis is the most common cause of spontaneous biloma formation. However they are reported to occur following acute cholecystitis, hepatic infarction/abscess, biliary tract neoplasm and biliary TB as well. It has been suggested that raised intraductal pressure secondary to obstructive pathology such as biliary tract calculi, stricture or neoplasm etc. may contribute in spontaneous biloma formation [4]. In our case the 'Biloma' was seen medial and inferior to left lobe of liver at subcapsular location. We failed to identify any direct contributing factor for biloma formation, so ours is probably a case of spontaneous subcapsular biloma, which is a very rare entity.

Bilomas usually present with pain in RUQ (right upper quadrant) and fullness in abdomen. They may or may not be associated with fever. They are usually seen in RUQ at subphrenic and subhepatic location [3]. Subcapsular bilomas are less commonly seen and they are reported following surgery and trauma [4-6].

Bilomas appear as a unilocular/mutilocular cystic lesion on ultrasonography. The lesions are usually anechoic, but they may contain moderate level echoes and show complex appearance,specially when infected. Ultrasound is also an important tool in providing guidance for diagnostic/therapeutic aspiration from biloma and for follow-up studies. CT scan is better in defining location,relation with adjacent structures and extent of the disease and is also useful in providing accurate guidance for drainage of the collection [7]. The appearance of biloma on USG and CT necessitates a differential diagnostic consideration of hepatic cyst, hepatic abscess,pseudopancreatic cyst,

seroma,hematoma and lymphocele [6]. Percutaneous aspiration under imaging guidance can be a problem-solving tool in certain cases.

(<4 cm) asymptomatic bilomas are managed without any intervention only follow-up is recommended [9]. However, most of the bilomas require some or the other form of therapeutic management. The conservative management of bilomas includes intravenous hydration and antibiotic therapy [7-9]. Large or symptomatic bilomas are usually managed by percutaneous drainage [9]. But some patients may require endoscopic nasobilliary drainage (ENBD) and endoscopic sphincterotomy or stenting to reduce the pressure in billiary channels. Surgery is reserved for select cases of persistent biliary leakage despite endoscopic therapy or to treat underlying disease [7, 9].

CONCLUSION

Spontaneous biloma formation is a very rare entity. A high index of clinical suspicion combined with relevant imaging findings and lack of other plausible etiologies is necessary for prompt recognition and its proper management.

Percutaneous treatment should be considered as the first-line option for patients with symptomatic spontaneous biloma. In cases of persistent bile leaks, endoscopic biliary drainage and endoscopic sphincterotomy with or without stent placement should be performed.

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