SPONTANEOUS BILOMA: REPORT OF TWO CASES WITH REVIEW OF THE LITERATURE

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ABSTRACT

Background Biloma is the collection of bile outside the biliary tree and can be intra or extrahepatic. It is most commonly iatrogenic, secondary to surgery or percutaneous transhepatic procedures or secondary to abdominal trauma. Spontaneous bilomas are very rarely described in the literature. Case reports Case 1: A 32-years-old female presented as lump abdomen in the epigastric region. Blood investigations were normal. Chest radiographs were unremarkable. Ultrasound examination revealed two cystic lesions about left lobe of liver and cholelithiasis with choledocholithiasis. Ultrasound-guided pigtail catheter drainage of the collection was done. ERCP failed to manage the choledocholithiasis. CBD exploration with choledocholithotomy and T-tube drainage and cholecystectomy was done. The postoperative period was uneventful, and the T-tube was removed on the 10th day after a normal T-tube cholangiogram. Case 2: A 32-year-old female admitted in an emergency with a complaint of acute pain in abdomen since two days. Ultrasound shows contracted gallbladder with stone along with focal collection in perihepatic and perigallbladder region extending into the subcutaneous plane with CBD stone. Ultrasound-guided pigtail catheter drainage of the collection was done. After five days she underwent ERCP by which CBD stones were cleared. Elective laparoscopic cholecystectomy was done. Conclusion A high index of clinical suspicion is necessary for prompt recognition and its proper management. Percutaneous treatment should be considered as the first-line option for patients with spontaneous biloma. In cases of persistent bile leaks, endoscopic biliary drainage and endoscopic sphincterotomy with or without stent placement should be performed. Surgery is now performed only in cases with a persistent bile leak or for treatment of an underlying disease.

KEYWORDS: chronic hepatitis B, reactivation of hepatitis B, HBV infection, autoimmune hepatitis, tenofovir, antivirals.

Introduction

The term ‘biloma’ was first coined by Gould and Patel in 1979 [1]. Kuligowska extended the term biloma to include intrahepatic as well as extrahepatic collections of bile [2]. 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 rupture of the biliary tree is rarely observed, sometimes being associated with choledocholithiasis [3, 4]. We report two cases of spontaneously formed bilomas secondary to extrahepatic biliary obstruction (EHBO), and we discuss how to diagnose and manage such patients, together with a review of
the related literature.

Case reports

Case 1
A 32-years-old female presented as an abdominal lump in outpatient department (OPD). Past medical and surgical history was not remarkable. At the time of examination, he was afebrile, hemodynamically stable. Abdominal examination revealed a lump in epigastric region and right upper quadrant.

Initial investigations revealed mild leukocytosis (16,300/mm³). Liver function tests, amylase and coagulation screen, were normal. An erect abdomen and chest radiographs were unremarkable. Ultrasound (USG) examination revealed two cystic lesions about left lobe of the liver. Other findings included cholelithiasis with choledocholithiasis. Computed tomography (CT) scan showed a subcapsular collection of 138 x 87 x 110 mm size anterior to the left lobe of liver and a subcapsular collection of 135 x 101 x 108 mm size posterior to the left lobe of the liver. On postcontrast scan, there was no enhancement. Additionally, there was cholelithiasis with choledocholithiasis and bilateral mild pleural collection. Ultrasound-guided pigtail catheter drainage of the collection was done, which removed about 400 ml and 350 ml of altered bile in the first 24 h after the procedure. The patient was kept on broad-spectrum antibiotics. Minimal drainage was noted after the first 48 h. Endoscopic retrograde cholangiopancreatography (ERCP) revealed a dilated common bile duct (CBD) and two stones (largest 20 mm) visible in the lumen. CBD exploration with choledolithotomy and T-tube drainage and cholecystectomy 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 10th day after a normal T-tube cholangiogram. On follow-up for three months, USG of the abdomen did not show any residual or recollection of bile. (Figure 1)

Case 2
Thirty-two-year-old female admitted in an emergency with a complaint of acute pain in abdomen since two days. History was insignificant. On examination, there was a lump in the right hypochondrium and right lumbar region of abdomen. Her investigations show leucocytosis (10,300) alkaline phosphatase is 569.5 U/L and serum bilirubin 2.1 mg/dl. Ultrasound shows contracted gallbladder with stone along with focal collection in perihepatic and perigallbladder region extending into the subcutaneous plane with CBD stone. Computed tomography scan showed well-defined hypodense fluid measuring 87 x 39 mm with enhancing rim and internal septations within the gallbladder fossa extending in the hepatorenal pouch and right flank region measuring 61 x 46 mm with choledocholithiasis. Ultrasound-guided pigtail catheter drainage of the collection was done in gallbladder fossa and right retroperitoneal region. Pus mixed with bile came out and gradually reduced in quantity. She was kept on broad-spectrum antibiotics. After five days she underwent ERCP by which CBD stones were cleared. The drain was removed on the 10th day, and the patient was sent home in stable condition. Elective laparoscopic cholecystectomy was done after 12 week followed by biliary stent removal. She was followed for three months, and USG does not show any collection of bile. (Figure 2)

Discussion
The most common underlying cause of spontaneous biloma is choledocholithiasis [5]. Other rare causes include bile duct tumours, hepatic infarction, abscess, obstructive jaundice, tuberculosis, and idiopathic [6]. The mechanism of spontaneous biloma formation is thought to be either the raised intraductal pressure secondary to extrahepatic biliary obstruction (EHBO)
due to stone, tumour, and spasm of the sphincter of Oddi, rupture of a cyst/diverticulum, or a focal liver infarction [5,6]. In our patients, EHBO with raised intraductal pressure was present due to choledocholithiasis in both cases.

Bilomas are less common and have been reported following surgery and trauma [6]. Spontaneous bilomas are seen very rarely.

The subcapsular location of bilomas in one patient 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 detergent activity of bile acids provokes chronic inflammation that, in turn, causes adhesions, leading to a possible loculated appearance of the collection [6]. Encapsulation of bile within the omentum and mesentery prevents generalised peritonitis in most cases.

Bilomas are more common in the right upper quadrant 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 [5,6]. The RUQ abdominal pain is the constant sign in the patient with biloma [7]. Ultrasound is sensitive for diagnosing bilomas, but the diagnosis of this complication is ideally facilitated by the use of CT [7]. CT scan is optimal for identifying and localising 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. Most bilomas have a CT number of less than 20 HU unless they are mixed with blood or exudates [8]. Biochemical and microbiological analysis of the fluid helps differentiation from pyogenic abscesses or other causes.

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.

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 gastrointestinal shadows can hide them, can be difficult to diagnosis [3].

USG scan can also guide proper drainage of the collections, as was done in our two patients.

Treatment for bilomas with a diameter of only a few centimetres is not always necessary; these lesions can be watched. However, most bilomas require treatment.

In the past, surgery was the primary approach to treatment [3]. 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 tumours, or when there is persistent active bile leakage in spite of percutaneous drainage [9].

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 collections and endoscopic stone retrieval.

Conclusion
Spontaneous biloma formation is a very rare entity. A gamut of endoscopic and radiological investigations often fails to localise the site of bile leak 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. Surgery is now performed only in cases with a persistent bile leak or for treatment of an underlying disease.

Authors’ Statements

Competing Interests

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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