Case reports of interest

Spontaneous idiopathic subcapsular biloma

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Abstract
Spontaneous biloma is an uncommon entity. We report a case of subcapsular biloma in an elderly patient with a non-obstructed biliary channel, without prior history of surgery, instrumentation, or trauma. Computed tomography (CT) and magnetic resonance imaging are described. We believe that this is the first reported case of spontaneous subcapsular biloma of idiopathic origin.

Key words Idiopathic · Spontaneous · Subcapsular biloma

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 iatrogenic (surgery and instrumentation) and trauma.

Nontraumatic bilomas are commonly referred to as spontaneous bilomas. Since 1979, some 27 cases of spontaneous bilomas have been reported. The most common underlying cause of spontaneous biloma is choledocholithiasis.2 Idiopathic biloma is a rare entity. Interventional radiology plays a pivotal role in the diagnosis and treatment of such cases.

Case report
An 80-year-old woman was admitted with a 2-month history of intermittent pain involving mainly the right upper quadrant (RUQ) of her abdomen. The present episode of pain had started 3 days previously and was associated with vomiting and constipation.

She was known to suffer from hypertension, hypothyroidism, and hypercholesterolemia. Past surgical history included appendectomy and right mastectomy for Paget’s disease 20 years earlier.

Although she was apyrexial and hemodynamically stable, she looked unwell. Abdominal examination was normal apart from minimal tenderness in the RUQ.

Initial investigations showed a raised white cell count (34.9 × 10^9/l), C-reactive protein (CRP; 492 mg/l), urea (10.7 mmol/l), and creatinine (203 µmol/l). Liver function tests, amylase, and coagulation screen were normal, as were her chest and abdominal plain films. The patient was commenced on intravenous antibiotics.

Ultrasound (US) scan showed a large subdiaphragmatic collection in the RUQ. The rest of the abdomen was normal. Computed tomography (CT) scan showed a large well-circumscribed subdiaphragmatic collection, 16 × 5 cm in size, over the right lobe of the liver, extending downwards on the lateral surface. Features were suggestive of a subcapsular collection (Fig. 1).

Bilious fluid was aspirated and an 8 French pigtail catheter was inserted into the collection under CT guidance. The aspirated fluid was confirmed to be bile by a dipstick technique. Fluid was sent for laboratory investigations. There was no growth on microbiology; cytology showed few red blood cells, many polymorphs and macrophages, and amorphous debris, but no malignant cells.

Magnetic resonance cholangiopancreatography (MRCP) revealed a normal biliary system and a collection extending around the lower margin of the right lobe of the liver towards the gallbladder, without any direct communication with the biliary channels (Fig. 2).

Following percutaneous drainage, the patient’s clinical condition improved dramatically. The drainage of bile ceased after a week. Follow-up US scan at this stage
showed almost complete resolution of the subdiaphragmatic collection. The indwelling catheter was removed and the patient was discharged from the hospital.

Her repeat US scans at 1 month and 3 months following discharge from hospital were normal.

Discussion

The meaning of the term “biloma”, initially coined to denote an encapsulated, extrahepatic collection of bile, has subsequently been modified to include intrahepatic bile collections located outside the bile ducts. Bilomas are most common after surgery, especially following cholecystectomy, instrumentations including percutaneous transhepatic cholangiography, liver biopsy, biliary drainage procedures, endoscopic retrograde cholangiopancreatography (ERCP), and trauma.

Spontaneous formation of biloma is now a recognized clinical entity. A review of reported cases of spontaneous biloma has revealed that this condition, similar to that in our patient, mainly affects people over 70 years of age. There is no sex preponderance and choledocholithiasis is the major causative factor. The other less common causes include malignancy of the biliary tree, hepatic infarction, and abscess.

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. We could not identify any cause or contributing factor for the spontaneous biloma in this 80-year-old woman. Imaging revealed a normal biliary tree. Previously, Caride and Gibson reported three cases of bilomas; in only one patient was the cause unknown.

The usual presentation in such cases 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.

Most of the collections are subphrenic and subhepatic. Subcapsular bilomas following surgery and trauma have been reported. We found one case report of spontaneous subcapsular biloma due to choledocholithiasis. Interestingly, in our patient, the spontaneous biloma was subcapsular without any contributing factor.

CT scan is optimal for identifying and localizing bilomas and showing their nature (unilocular or septate), distribution, and regional anatomy. Differential diagnoses include 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 Hounsfield units (H) unless they are mixed with blood or exudate. In our patient, the CT number of the collection was 5 H.

MR scan complements the CT in making a diagnosis. It also helps to differentiate biloma from subacute hematoma; this is important, as drainage is not required for the latter. MRCP is an important investigation to identify the etiology. All these investigations in our

Fig. 1. Computed tomography (CT) scan with intravenous contrast showing the subcapsular biloma and normal gallbladder (arrows)

Fig. 2. Magnetic resonance cholangiopancreatogram image showing the subcapsular biloma (large arrow) and the normal biliary tree (small arrow)