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Case Report

Spontaneous biloma manifesting with massive upper gastrointestinal bleed: case report

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ABSTRACT

Spontaneous biloma is a rare non-traumatic lesion in which the extrahepatic or intrahepatic bile duct perforates spontaneously with no discernable cause. We present a case of spontaneous biloma in a 68 years old male patient with cholelithiasis and concurrent choledocholithiasis, who presented with acute severe cholangitis. There was no history of abdominal surgery, instrumentation, or trauma in the past. Initial ultrasonography (USG) abdomen showed central intrahepatic dilation, cholelithiasis, and choledocholithiasis. A sizeable hypoechoic lesion with few internal septations was present adjacent to the left lobe of the liver, with USG suspicion of a hydatid cyst. The lesion was abutting and displacing the stomach anteromedially. The patient had an acute renal failure with hemodynamic instability at presentation. The patient was managed with emergency endoscopic drainage under ultrasound guidance and improved after that. On the fourth day, after endoscopic drainage, the patient had a massive upper gastrointestinal (UGI) bleed due to erosion and pressure necrosis of the gastric wall due to biloma. The patient was managed successfully with sleeve gastrectomy and de-roofing of the cyst.

Keywords: Biloma, Spontaneous bile leak, ERCP, Choledocholithiasis, UGI bleed

INTRODUCTION

Nontraumatic bilomas are commonly referred to as spontaneous bilomas. The most common underlying cause of spontaneous biloma is choledocholithiasis.[1]

We report an interesting case of spontaneous biloma, mimicking hydatid cyst and manifesting with a massive UGI bleed in a patient of choledocholithiasis with acute severe cholangitis.

CASE REPORT

A 68 male presented with symptoms of obstructive jaundice of 01-month duration and high-grade fever associated with chills, rigors, and decreased urine output for the last three days. There was no history of abdominal surgery, instrumentation, or trauma in the past.

On examination, the patient was febrile and deeply icteric. He was hypotensive; his blood pressure was 80/60 mm of Hg, and his pulse rate was 132/minute. On abdominal examination was unremarkable.

The laboratory workup was as follows: Hemoglobin (Hb), 11.2 gm/dl (13-16); white blood cell count, 16100/cubic millimetre (4000-11,000); total bilirubin, 13.2 mg/dl (0.2-1.2), conjugated bilirubin, 7.49 mg/dl (0-0.3); aspartate aminotransferase, 380 IU/L (02-41); alanine aminotransferase, 457 IU/L (02-40); alkaline phosphatase, 1410 IU/L (42-128), serum urea 147 mg/dl (10-50), creatinine 3.09 mg/dl (0.5-1.2), INR 1.7. The viral and hydatid serology were negative.

There was central intrahepatic dilation on the ultrasonography (USG) abdomen. A 12 mm stone was present in the common bile duct with distal cut-off and

multiple calculi in the gall bladder (GB). A large, hypoechoic lesion with few internal septations was present adjacent to the left lobe of the liver, measuring 7×7 cm, with USG suspicion of a hydatid cyst.

After optimising and correcting coagulopathy, the patient was assessed for percutaneous biliary drainage (PBD). There was no peripheral duct suitable for PBD.

Endoscopic retrograde cholangiopancreatography (ERCP) under USG guidance was done. On CBD cannulation, infected bile was aspirated, and a 10 Fr×05 cm double pigtail (DPT) stent was placed. In the side-viewing endoscope, a bulge was noted in the posterior wall of the stomach, and the gastric mucosa was reported as normal.

The patient improved following endoscopic drainage, and organ failure started resolving. The patient developed mild acute pancreatitis post-ERCP, managed conservatively.

The patient developed melaena on the fourth day after ERCP, and his Hb dropped to 7.6 gm/dl from 11.2 gm/dl. On contrast-enhanced CT scan of the abdomen with angiography, there was a well-defined, peripherally enhancing lesion measuring 11.7×8.8×8.2 cm in the left subphrenic location, displacing the fundus and the greater curvature of the stomach anteromedially (Figure 1 A-F).

On UGI endoscopy, there was an active ooze from the posterior gastric wall near the fundus, and the stomach was full of blood clots. The oesophagus was normal, and no varices were seen. The Hb of the patient dropped further to 3.4 gm/dl, and the patient was scheduled for emergency surgery.

On exploring the abdomen, changes of recent acute pancreatitis were noted. A cystic lesion in the left subphrenic location was present, abutting the left lobe of the liver, the posterior wall of the stomach near the fundus, and the greater curvature. The hepatoduodenal ligament was frozen. The fundus and greater curvature, adherent to cyst, were inflamed and friable, and on gastrotomy, the stomach was full of blood clots (Figure 2 A). De-roofing of the cyst was done, and bile mixed contents, mimicking hydatid membranes, were evacuated (Figure 2 B and C).

A sleeve gastrectomy of the stomach was done to excise the unhealthy gastric wall, and a feeding jejunostomy was added for early feeding (Figure 2 D). The patient recovered well, was discharged on the seventh postoperative day, and did well on follow-up.

The histopathology report showed transmural necrosis of the stomach wall. The cyst wall and contents showed necroinflammatory infiltrate admixed with biliophages. No granuloma, hydatid membrane, or hooklets were seen.

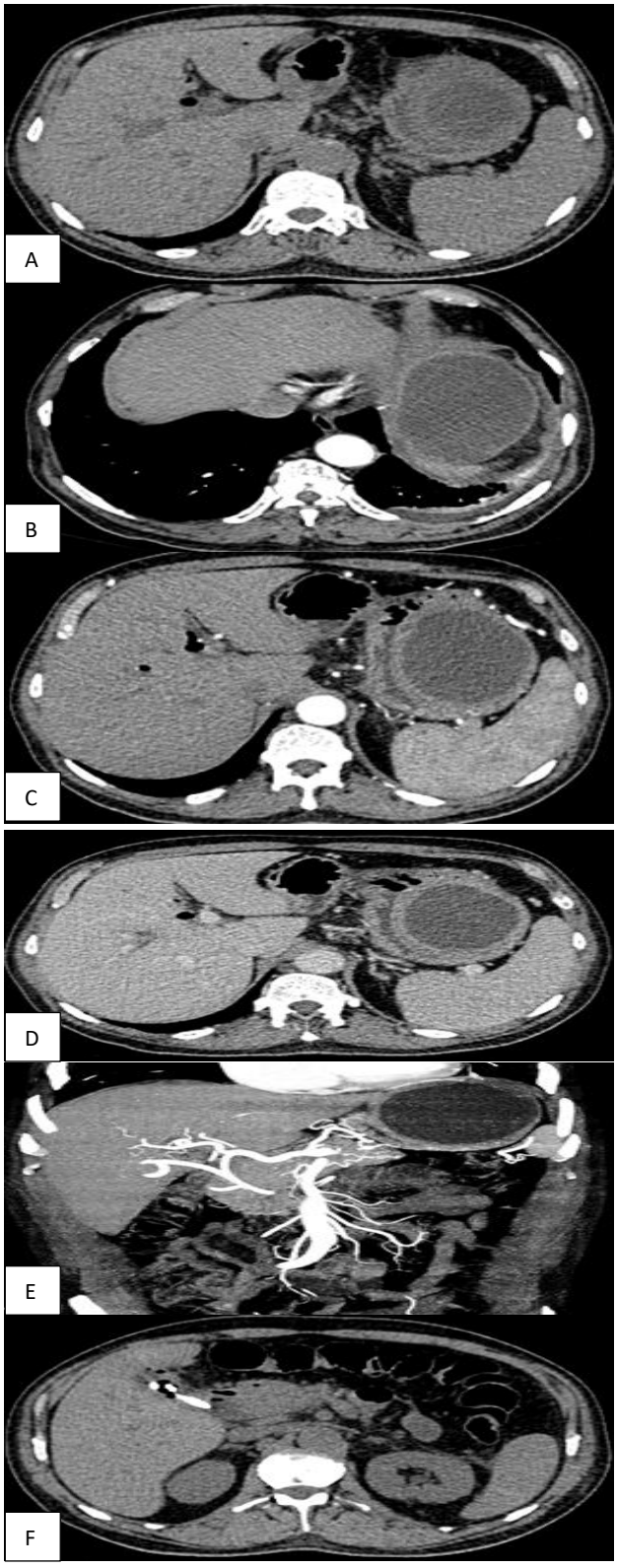

Figure 1 (A-F): CECT abdomen-axial images of abdomen; NCCT, arterial phase, venous phase and sagittal reconstruction images of well-defined, peripherally enhancing, hypodense lesion in left subphrenic region, displacing greater curvature of stomach with pneumobilia. NCCT axial image of CBD stent *in situ*. Central intrahepatic bile duct dilation with pneumobilia is evident if A, C, D, and E.

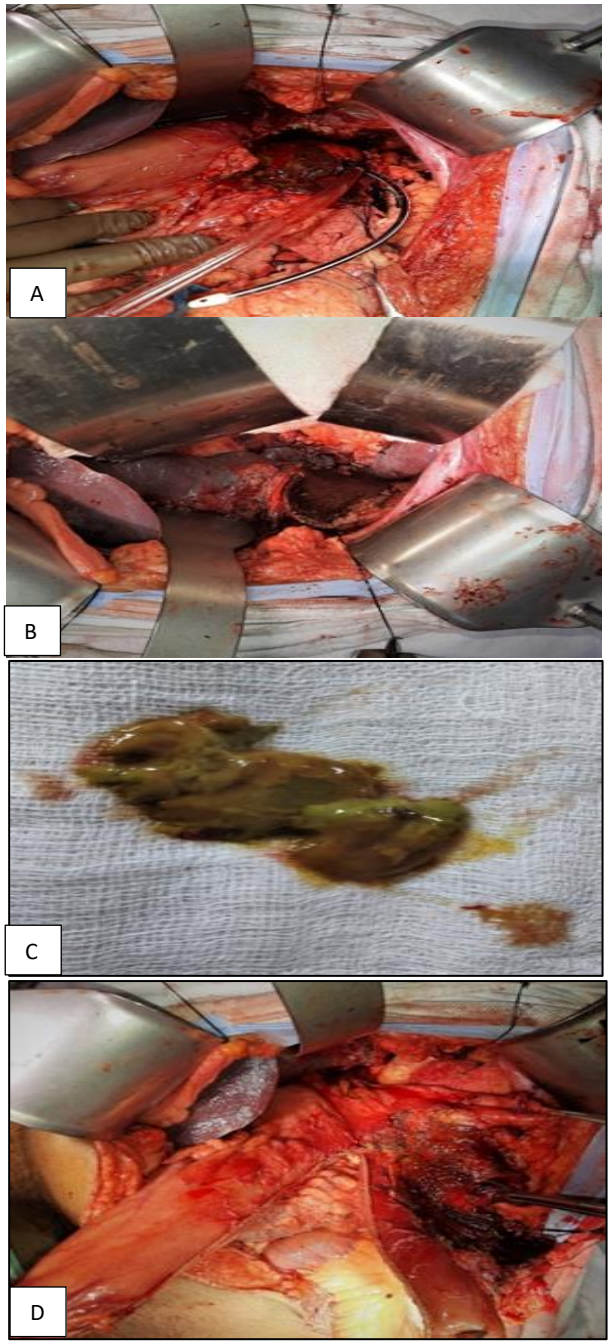

Figure 2 (A-D): Unhealthy, inflamed gastric wall, Ryle's tube coming out of necrosed part of the stomach near greater curvature and the stomach was full of blood clots. De-roofed cystic lesion. The lesion was abutting the left lobe of the liver, GE junction, and stomach fundus. Bile mixed contents, mimicking hydatid membrane evacuated from cystic lesion. Sleeve gastrectomy done with the help of linear stapler and stapler line reinforced with suture.

DISCUSSION

In 1997, Fujiwara et al reported spontaneous biloma in 73 years old female without any identifiable cause.[1] The authors also reviewed the details of all 25 cases of spontaneous biloma reported in the literature from 1979-1997.[1]

Bilomas are most common after surgery, especially following cholecystectomy, instrumentations including percutaneous transhepatic cholangiography, liver biopsy, biliary drainage procedures, ERCP, and trauma.[2]

The precise mechanism underlying spontaneous biloma development remains unclear. The suggested contributing factors include increased intraductal pressure caused by an obstruction that results from a stone, tumour, or sphincter of Oddi spasm.[1,3]

The clinical presentation of a biloma is variable, ranging from an incidental finding on imaging in an otherwise asymptomatic patient to abdominal fullness, pain, fever, and jaundice, to very rarely peritonitis without fever.[4]

Our case had spontaneous biloma at presentation to the hospital, and there was no history of any instrumentation. The lesion was thought to be an incidental USG finding of an asymptomatic hydatid cyst.

Treatment for spontaneous bilomas is mainly non-operative, with percutaneous drainage and endoscopic procedures.[5]

The percutaneous drainage of the biloma may have averted the need for emergency surgery, but since it was masquerading as a hydatid cyst, it was not attempted in this case. Further, a cholangiogram could have demonstrated a connection between the cavity and biliary system, but bedside ERCP under USG guidance was done in this case, and the same was missed.

CONCLUSION

The biloma presenting with massive UGI bleed is an unusual occurrence. The biloma should be in the differential diagnosis of cystic lesions in the upper abdomen, especially in a patient with biliary obstruction.

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