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PICTURES IN DIGESTIVE PATHOLOGY

Biliary peritonitis secondary to spontaneous rupture of hepatic mesenchymal hamartoma

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INTRODUCTION

The biliary hamartoma usually courses asymptomatic; we present an unusual clinical manifestation.

CASE REPORT

This is a 77 year old female with no medical history who presented to the emergency room with 48 hours of right hypochondrium abdominal pain, associated to conjunctive jaundice. The blood test showed elevated bilirubin and C reactive protein test. CT scan revealed gallbladder hydrops, dilated left intrahepatic bile duct, common bile duct (CBD) of 8 mm and left hepatic lobe atrophy (Fig. 1). The patient was admitted with the diagnosis of mild cholangitis and started antibiotic therapy. Cholangio-MRI showed cholelithiasis, distal choledocholithiasis with bile duct dilatation, left hepatic lobe atrophy with a dilated segmental bile duct of chronic aspect, right hepatic hypertrophy and millimeter diffuse cystic lesions (Fig. 2). Suddenly, her condition deteriorated associated with a diffuse peritonitis. She underwent a right subcostal laparotomy on

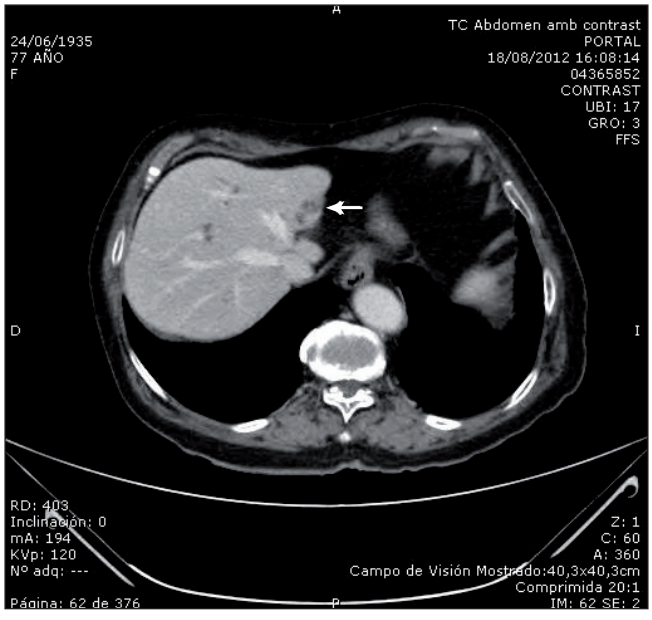

Fig. 1. Abdominal CT scan. Left hepatic lobe atrophy, dilated bile duct at this level (arrow).

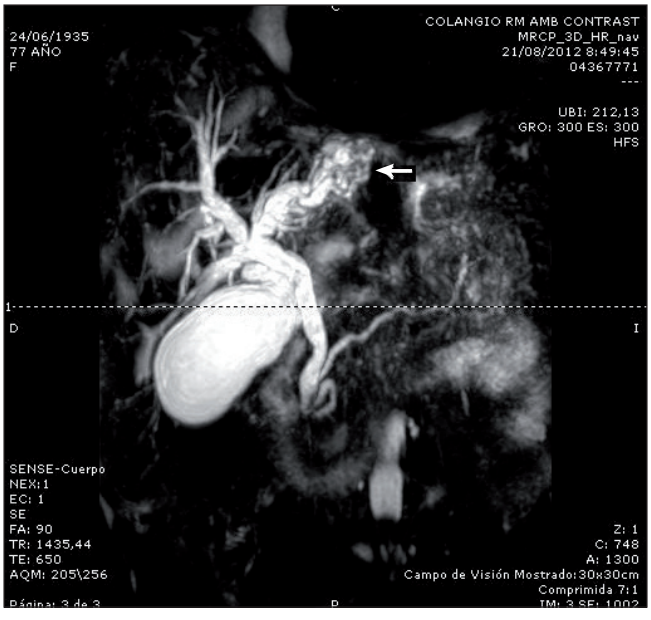

Fig. 2. Cholangio-MRI. Beaded dilatation of left segmental bile duct of chronic aspect (arrow).

emergency basis showing a choleperitoneum, normal gallbladder and the left hepatic lobe atrophy was confirmed. A perforated fibrous-cystic complex with a bile leak was found (Fig. 3). Intraoperative cholangiography illustrated the choledocholithiasis and revealed contrast leakage in the left hepatic lobe lateral segments (Fig. 4). Cholecystectomy, atrophic liver segment resection, choledochotomy with stone extraction and choledochorrhaphy over Kehr were performed as surgical treatment. She had an uneventful postoperative course. The histologic diagnosis was: Multicystic biliary hamartoma of 1.3 cm on its larger dimension, associated with acute inflammatory process (Fig. 5).

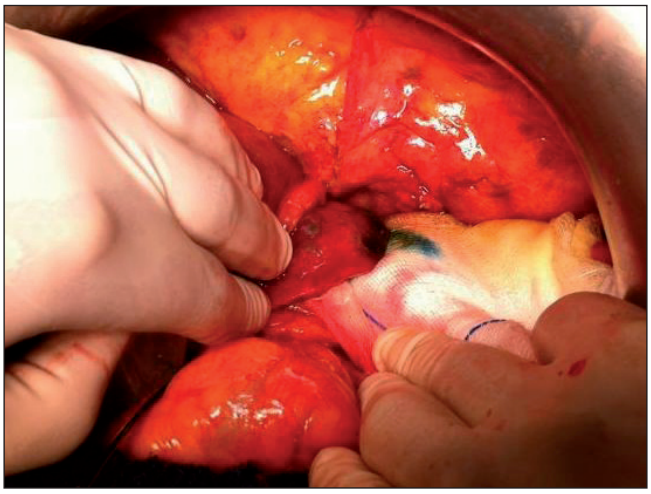

Fig. 3. Intraoperative image. Bile leak localized at the perforation level in liver´s atrophic portion manifested after cystic duct injection of methylene blue.

DISCUSSION

The biliary mesenchymal hamartoma is a benign tumor arising from the portal triad mesenchyme. Usually presents with multiple cystic liver lesions. It is uncommon in adults. It is usually asymptomatic and it may produce local symptoms on large cysts cases. Spontaneous bile duct perforation is extremely rare and up to date we have not found any cases reported associated with biliary hamartoma. Increased CBD pressure due to impacted stones is postulated as perforation cause.

Fig. 4. Cholangiography. Extravasation of contrast.

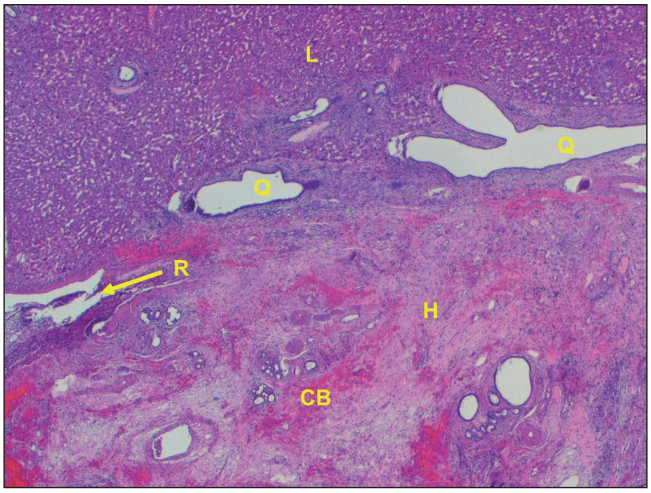

Fig. 5. Histological study. Multicystic biliary hamartoma: h/e 200 field. H: Area of hamartoma. L: Area of normal liver. Q: Biliary cysts. CB: Bile ducts. A: Focus hamartoma rupture with fibrin deposition and acute inflammatory component.

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