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Hepatolithiasis with secondary cholangitis and supernumerary left hepatic lobe

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ABSTRACT

We report the case of a 31-year old woman with recurrent cholangitis secondary to hepatolithiasis. The stones were composed of calcium bilirrubinate. The patient also had a supernumerary hepatic lobe connected to the inferior aspect of the segment III of the liver. The role of the supernumerary hepatic lobe in the development of hepatolithiasis is unclear and may be coincidental.

Key words. Lithiasis. Cholangitis. Extrahepatic bile ducts. Gallbladder.

INTRODUCTION

To our knowledge the association of hepatic supernumerary lobe and hepatolithiasis has not been reported and may be coincidental.[1] In hepatolithiasis, the stones are usually located proximal to the confluence of the right and left hepatic ducts.[2] Two types of intrahepatic stones have been described: one consists of calcium bilirrubinate (brown pigment and black-coloured mixed stones) and the other of pure cholesterol.[3] Hepatolithiasis is characterized by a high rate of treatment failure and recurrence.[4]

CLINICAL CASE

A 31-year-old woman was admitted with a history of multiple hospitalizations. The first hospitalization was characterized by abdominal pain, fever and jaundice. A clinical diagnosis of choledocolithiasis and cholelithiasis was performed, and the patient was treated with a therapeutic endoscopic retrograde cholangiopancreatography (ERCP) and laparoscopic cholecystectomy.

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During the last hospitalization, she developed intermittent abdominal pain without fever or jaundice. On physical examination she had abdominal tenderness in the right upper abdominal quadrant. The laboratory data showed normal liver enzymes, bilirubin and coagulation profile. There was no leukocytosis or anemia. Hepatic magnetic resonance images (MRI) revealed a supernumerary lobe connected to the segment III of the liver (Figure 1A). In addition, there was hepatolithiasis with intrahepatic bile duct dilatation in the left lobe. Moreover, the magnetic cholangioresonance showed a vertical disposition of the intrahepatic bile ducts of the supernumerary lobe and hepatolithiasis of the left lobe.

Based on clinical findings and the imaging studies a subtotal left hepatic lobectomy was performed.

Surgical findings

At the operation, the gallbladder was absent and its bed was found at the left of a prominent round ligament. The supernumerary lobe was dissected with ease from the remaining liver. Individual ligations of the portal vein and artery as well as a venous drainage were performed. The aberrant bile duct of the supernumerary lobe had a right angle insertion in the common hepatic duct. When it was removed a large defect was observed in the area of the common hepatic duct, and a conventional Roux-Y hepatojejunostomy was performed. There were no post-opera-

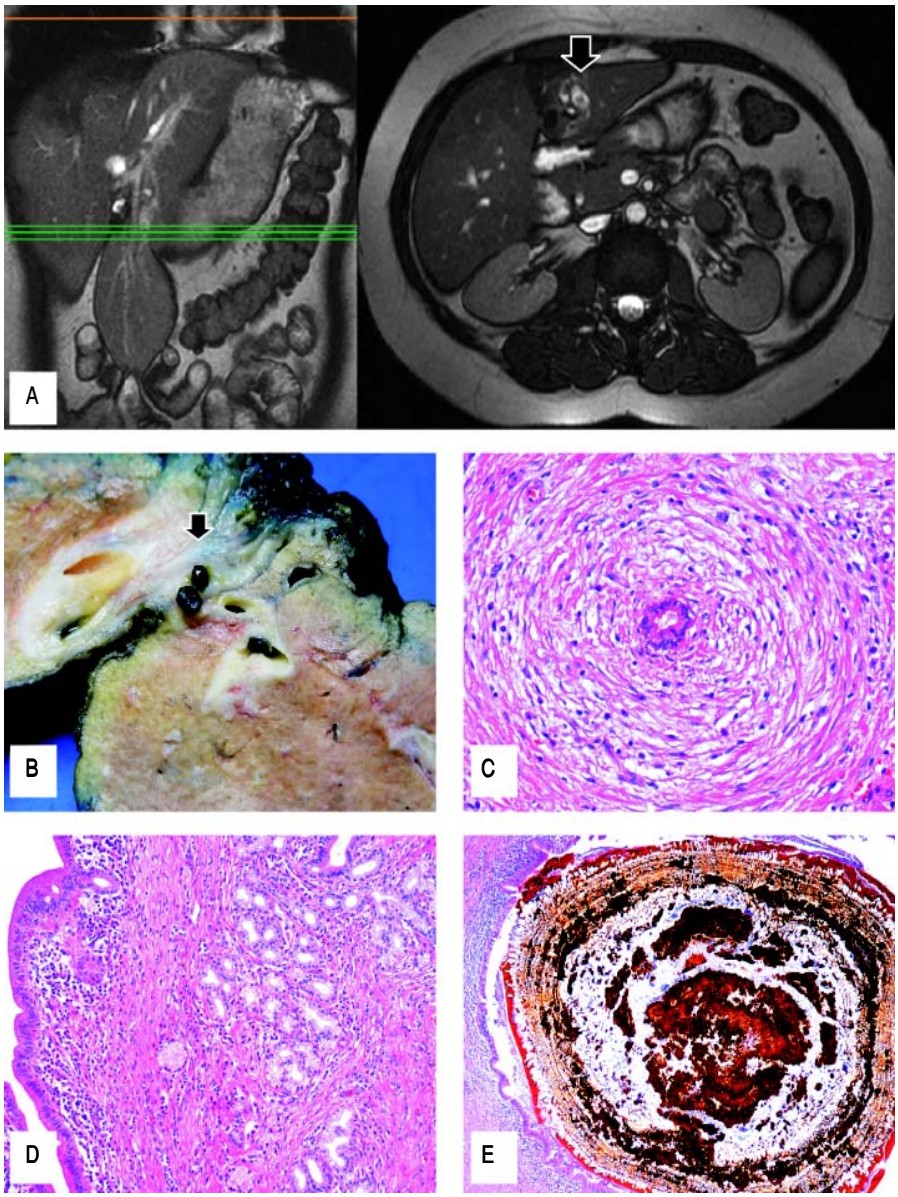

Figure 1. A. *The quality of the figure is low. Hepatic MRI in T2, the coronal cut (right) shows a supernumerary lobe in the segment III (left hepatic lobe) with intrahepatic dilatation and hepatolithiasis (arrow).* ***B.*** *The hepatic lobectomy specimen showed dilated bile ducts with fibrosis of their walls and intraluminal brown and dark-green stones (arrow). The intrahepatic bile ducts showed concentric fibrosis (**C**) and hyperplasia of metaplastic intramural biliary glands (**D**) (H&E stain, 200X), similar to those of end-stage PSC lesions.* ***E.*** *An intraluminal calcium bilirrubinate stone is shown (H&E stain, 200X).*

tive complications and the patient was discharged 5 days later.

Gross and microscopic pathology

The hepatic lobectomy specimen consisted of two irregular fragments of hepatic parenchyma that measured 13 x 4.5 x 3.2 cm and 10 x 5 x 3.5 cm. The multiple cut sections showed dilated intrahepatic bile ducts with ductal fibrosis. In addition, there were numerous brown and dark-green stones that measured 0.4 cm in greatest dimension (Figure 1B). The cut surface of the hepatic parenchyma was normal.

Microscopically, the intrahepatic bile ducts were dilated and showed periductal concentric fibrosis (Figure 1C). There were also hyperplasia of metaplastic intramural biliary glands (Figure 1D) and nerve trunks, similar to those of the end-stage primary sclerosing cholangitis (PSC) lesions.[5] Moreover, the bile duct walls showed multiple foci of both acute and chronic inflammatory cells associated with intraluminal calcium bilirrubinate stones (Figure 1E). In some areas, the portal spaces showed inflammatory microabscesses and lymphoid follicles with germinal centers. The liver parenchyma was normal.

DISCUSSION

Hepatic supernumerary lobes are relatively common, mainly in adult women, and are connected to the inferior surface of the liver.[1] Hepatic supernumerary lobes are connected by a mesentery containing branches of the portal vein, hepatic vein and hepatic artery, and a bile duct.[1] Supernumerary lobes may rarely require surgical treatment because of their large size, torsion of the pedicle, or the presence of other associated defects.

Hepatolithiasis is most common in East Asian countries, where the prevalence has been reported to be 20% of all patients undergoing surgery for gallstone disease.[2,6] In Western countries, including Mexico, the incidence is less than 1% and is generally thought to be secondary to stones originating in the gallbladder or primarily resulting from benign strictures, primary sclerosing cholangitis, choledochal cysts or malignant biliary tumors. In the East it is regarded as a separate entity altogether. The majority of cases of hepatolithiasis are associated with recurrent pyogenic cholangitis, mainly in regions of parasitic infestation. Hepatolithiasis frequently recurs, leading to cirrhosis and liver failure from long-term cholangitis.[7-8] The ultimate goal of treatment is the complete removal of stones and possible resolution of the biliary stricture.[9-10]

The etiology of hepatolithiasis in our patient is unclear. It is possible that the gallstones previously removed and the extrahepatic bile duct stones may have played a role in the development of intrahepatic stones.

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