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IMAGE | PANCREAS

Jack in the Duct: A Case of Common Biliary Duct Schwannoma

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CASE REPORT

A 46-year-old woman was admitted with a history of painless, intermittent, self-resolving, cholestatic jaundice, without preceding prodrome for the past 1 year. She had 3 similar episodes without any significant past medical history. There was no history of significant weight loss, anorexia, hepatic decompensation, or any surgery in the past. Laboratory investigations showed a normal hemogram, conjugated hyperbilirubinemia with total bilirubin of 12 mg/dL and direct bilirubin of 9.6 mg/dL, normal amino-transferases, of 38 IU/L, elevated alkaline phosphatase (1,123 IU/L), and an albumin level of 3.4 g/dL with a gamma-glutamyl transferase (GGT) of 63 U/L. Hepatitis A, B, and C virus serology results were negative.

Ultrasonography of the abdomen showed an 8 mm dilatation of the mid- and proximal common biliary duct (CBD) with intra-hepatic biliary radicle dilatation (IHBRD). A contrast-enhanced computed tomography scan of the abdomen revealed an ill-defined lesion in the lower CBD, with IHBRD. Magnetic resonance cholangiopancreatography demonstrated IHBRD and T2 hyperintense focal cystic bulbous dilatation of the left hepatic duct (Figure 1).

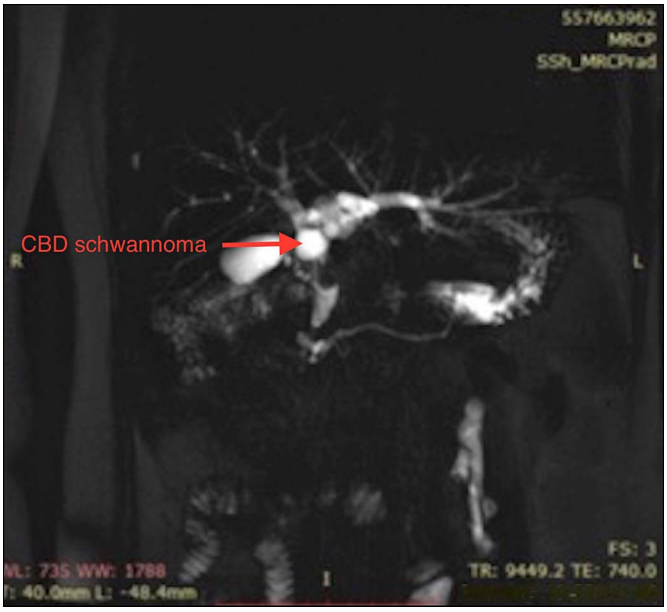

Figure 1. Magnetic resonance cholangiopancreatography showing common biliary duct mass.

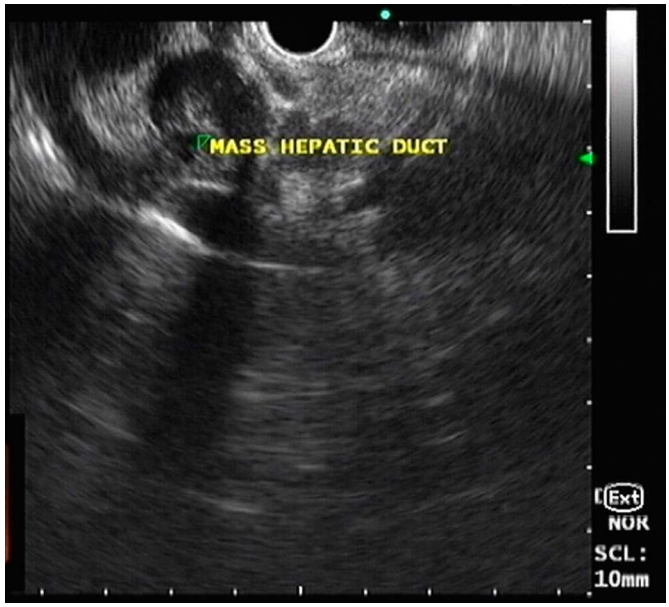

Figure 2. Endoscopic ultrasound image showing a mass in the hepatic duct.

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Figure 3. Cholangioscopy image of polyp.

Endoscopic ultrasound was suggestive of an eccentric globular mass with dilated CBD and IHBRD (Figure 2). Endoscopic retrograde cholangiopancreatography revealed an ulcerated, pinkish fleshy mass hanging out of papilla intermittently. Cholangiogram demonstrated dilated mid- and proximal CBD with IHBRD with rounded filling defect in the left hepatic duct. Spyglass cholangioscopy (Boston Scientific, Marlborough, MA) showed a polypoidal lesion with a smooth surface and ulcerated stalk attached at the hilum (Figure 3). Biopsies were taken during cholangioscopy using SpyBite forceps (Boston Scientific, Marlborough, MA). CBD stenting was performed.

Biopsy showed fascicles of benign spindle cells mixed with sparse inflammatory cells. Immunohistochemistry showed S100 positivity suggestive of Schwannoma of the biliary tract (Figure 4). Our patient did not consent for definitive surgery and is currently asymptomatic. CBD Schwannoma arises from sympathetic and parasympathetic nerve fibers around the wall of the CBD. Treatment is resection of CBD and hepaticojejunostomy.[1] There are only 18 reported cases until now, which showed female predominance and an average age of 43 years. The most common symptom at presentation was jaundice.[2]

Almost all the reported cases showed involvement of the extrahepatic bile ducts. In a similar case, reported by Madhusudan et al, it was shown to be involving both intrahepatic and extrahepatic bile ducts.[3] Our case is a unique one as it is arising from the isolated intrahepatic bile duct, pedunculated, intermittently prolapsing and presenting as intermittent obstructive jaundice. Our case has no other features suggestive of neurofibromatosis. Keeping the possibility of such tumor and timely referral after endoscopic retrograde cholangiopancreatography is the key to diagnosis and preventing

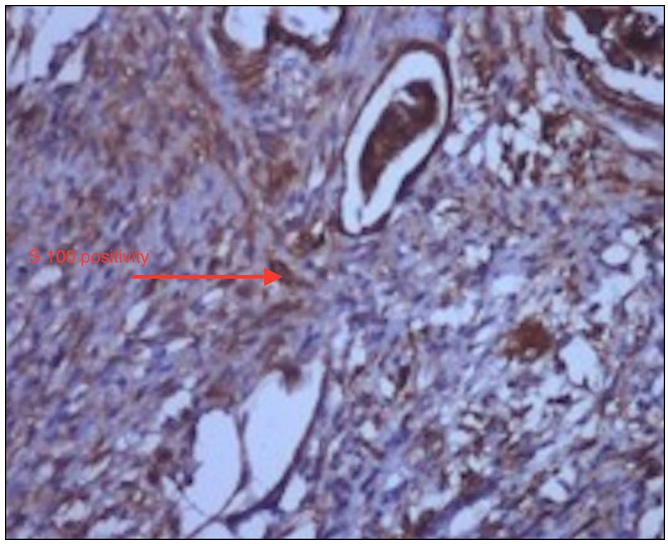

Figure 4. Immunohistochemistry showing S100 positivity of the spindle cell tumor.

surgery. Also use of cholangioscopy-guided biopsy was fundamental in preoperative diagnosis of the lesion, unlike previous cases where the diagnosis was based on postsurgical histopathology.

DISCLOSURES

Author contributions: All authors contributed equally to this manuscript. KM Kolhe is the article guarantor.

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Informed consent was obtained for this case report.

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