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Case Report

Intraductal Papillary Neoplasm of the Biliary Tract: A Rare Pre-Malignant Diagnosis

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INTRODUCTION

Intraductal papillary neoplasm of the biliary tract (IPNB) is a rare premalignant lesion of the biliary tree and a precursor of cholangiocarcinoma.[1] It is analogous to intraductal papillary mucinous neoplasm of the pancreas (IPMN-P), which has established guidelines for standardized surveillance.[2] Compared with IPMN-P, IPNB carries a higher risk of progression to invasive cholangiocarcinoma.[3,4]

IPNB arises from the bile duct epithelium and exhibits papillary or villous morphology. Imaging typically demonstrates ductal dilatation with papillary frond-like filling defects or intraductal masses.[5-7] Magnetic resonance imaging (MRI) is important for identifying malignant transformation, with features such as mass-like ductal thickening, restricted diffusion, and progressive delayed enhancement.[8] MRI combined with endoscopic retrograde cholangiopancreatography (ERCP) and tissue sampling is recommended for early diagnosis and surgical resection.[1]

Diagnosis remains challenging due to nonspecific clinical features and overlap with other biliary disease. Recognition of the characteristic imaging findings is essential for timely diagnosis and management.

CASE REPORT

A 46-year-old woman presented to the emergency department with a one-week history of right flank pain, nausea, and vomiting. She had recently completed a course of antibiotics for a urinary tract infection. On physical examination, she appeared ill and had right upper quadrant tenderness. Laboratory evaluation revealed leukocytosis and elevated alkaline phosphatase, while serum lipase and urinalysis were within normal limits.

Initial contrast-enhanced computed tomography (CT) demonstrated marked intra- and extra-hepatic biliary ductal dilatation, most pronounced in the posterior right hepatic lobe (Figure 1). Same-day MRI with magnetic resonance cholangiopancreatography (MRCP) confirmed the dilatation but revealed no discrete intraductal or obstructing mass (Figure 2A, B). Given these findings, the patient was evaluated by gastroenterology and interventional radiology. ERCP demonstrated a dilated common bile duct (Figure 3A). A subsequent transhepatic cholangiogram with biliary drain placement was performed (Figure 3B). Cytology from brushings revealed high-grade dysplasia, raising concern for invasive carcinoma.

The patient ultimately underwent right hepatectomy with Roux-en-Y hepaticojejunostomy. Final pathology confirmed IPNB with high-grade dysplasia and multiple microscopic foci of periductal invasive carcinoma. Her postoperative course was complicated, requiring prolonged hospitalization, after which she initiated chemotherapy for IPNB with invasive carcinoma.

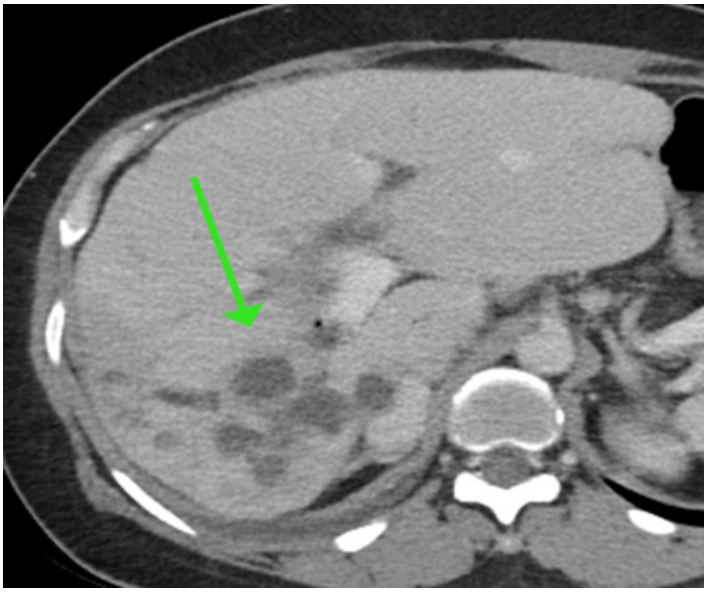

Figure 1. Axial CT image of the abdomen with contrast demonstrates marked biliary ductal dilatation (arrow) in the posterior right hepatic lobe without discrete obstructing or intraductal mass from IPNB.

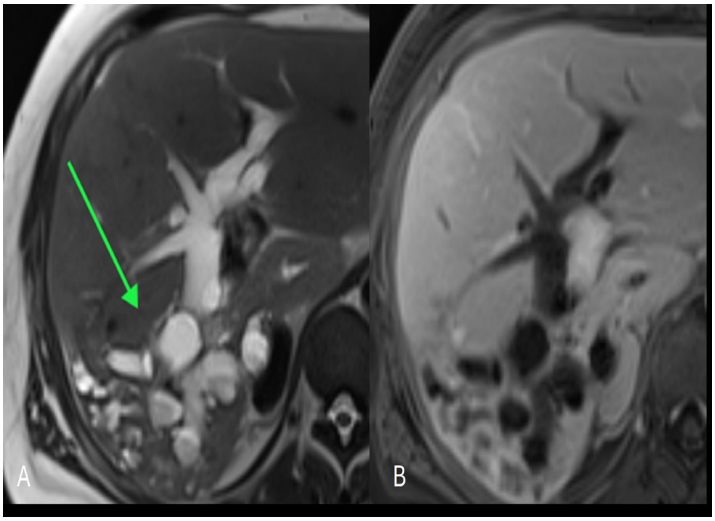

Figure 2. MRI abdomen with and without contrast. (A) Axial T2 image demonstrates marked intrahepatic biliary ductal dilatation (arrow) in the posterior right hepatic lobe from IPNB; (B) Axial postcontrast T1 fat saturated image without enhancing intraductal mass or filling defect.

DISCUSSION

IPNB is a neoplasm arising from the bile duct epithelium with papillary or villous morphology.[1,5] It is a slow-growing lesion that can undergo malignant transformation to cholangiocarcinoma and is histologically classified as low- to intermediate-grade dysplasia, high-grade dysplasia, or IPNB with associated invasive carcinoma.[9] Patients may present with abdominal pain, cholangitis, or obstructive jaundice with elevated alkaline phosphatase.[1,7,9] Although more common in Asian populations, IPNB accounts for only 7-11% of bile duct tumors in Western countries, with risks

factors including primary sclerosing cholangitis, choledochal cysts, and Gardner syndrome.[1]

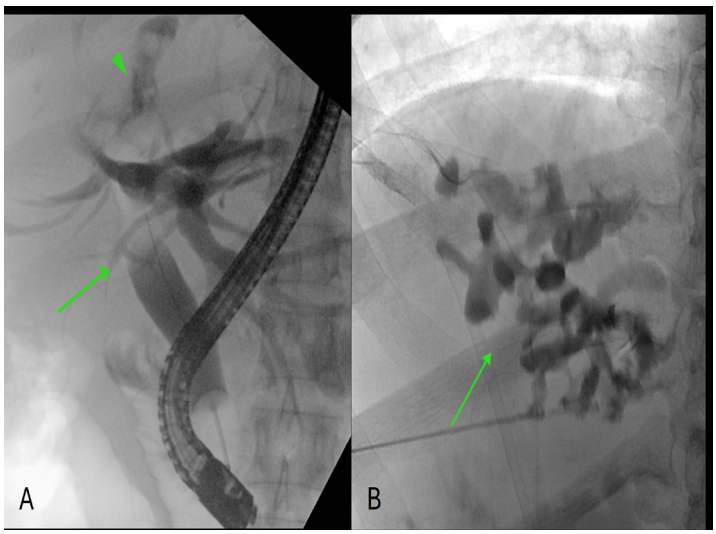

Figure 3. (A) ERCP image demonstrates marked extrahepatic (arrow) and intrahepatic (arrowhead) biliary ductal dilatation from IPNB; (B) Percutaneous right transhepatic cholangiogram with marked intrahepatic biliary ductal dilatation (arrow) without discrete mass from IPNB.

Imaging findings vary depending on mucin production and intraductal tumor size, location, and morphology. Ultrasound and CT often reveal nonspecific biliary ductal dilatation with or without intraductal masses or papillary filling defects, while MRI/MRCP is more sensitive for detection.[9,10] ERCP frequently is used in conjunction with cross-sectional imaging to evaluate mucin production and obtain tissue samples. Four radiologic patterns of IPNB have been described: (1) intraductal mass with aneurysmal bile duct dilatation both upstream and downstream from the lesion; (2) intraductal mass with upstream dilatation; (3) multiple intraductal masses with aneurysmal upstream dilatation and variable downstream dilatation; and (4) diffuse bile duct dilatation without a discrete mass.[1,5,9,11] Diffusion-weighted imaging further enhances the detection of invasive carcinoma, particularly with periductal infiltration.[8,10]

In our case, imaging demonstrated diffuse biliary ductal dilatation without a discrete mass, and pathology confirmed IPNB with microscopic foci of invasive carcinoma. Although microscopic invasive carcinoma was found after tissue sampling, no imaging findings suggestive of cholangiocarcinoma were identified on MRI. Even without overt malignant imaging features, IPNB is a premalignant lesion that requires close surveillance with MRI/MRCP and ERCP with tissue sampling. Diagnosis often is challenging due to its nonspecific presentation and overlap with other biliary diseases, such as ascending cholangitis.

CONCLUSIONS

In summary, IPNB is the rare biliary counterpart of the more common pancreatic IPMN. Because patients with IPNB are at increased risk of malignant transformation to invasive cholangiocarcinoma, recognition of its imaging features is important for early diagnosis and timely treatment.

ARTICLE INFORMATION

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