Intraductal papillary neoplasm of the bile duct is a precursor of invasive cholangiocarcinoma [1]. Early identification and intervention may improve the poor prognosis of this disease, but diagnosis by classical imaging is often difficult [2]. Also, as surgery is the therapy of choice, a precise pre-operative evaluation is necessary [3, 4]. Cholangioscopy may contribute to the diagnostic work-up.

A 40-year-old man without medical history presented with acute epigastric pain, jaundice, and elevated liver enzymes. Abdominal ultrasound showed intrahepatic and extrahepatic biliary ductal dilation without gallstones (▶Fig. 1). Echo-endoscopic ultrasound confirmed a diffusely dilated biliary tree with discrete hyperechogenic intraluminal material in the common bile duct, proximal to the cystic duct (▶Fig. 2). Endoscopic retrograde cholangiopancreatography with papillotomy was performed, which revealed spontaneous evacuation of mucin, which was further evacuated by balloon extraction (▶Fig. 3). Brush cytology was performed, and a 9-cm 10-Fr straight plastic stent was placed to ensure biliary drainage. Brush cytology revealed specific inflammation without dysplasia.

Additional peroral SpyGlass cholangioscopy (Boston Scientific, Marlborough, Massachusetts, USA) confirmed a lesion with papillary protrusions just below the liver hilum and in the distal right hepatic duct. The left intrahepatic ductal system was normal (▶Video 1). Biopsy specimens (SpyBite biopsy forceps, Boston Scientific) confirmed the diagnosis of multifocal intraductal papillary neoplasm of the bile duct with low-grade intraepithelial neoplasia (▶Fig. 4).

After multidisciplinary evaluation, a right hepatectomy with hepaticojejunostomy was performed. Pathologic examination of the resected specimen showed intraductal papillary neoplasm of the bile duct of the intestinal type (MUC1 negative, diffuse MUC2 positive, focal MUC5AC positive, limited MUC6 positive) with low-grade dysplasia, R0-resection (▶Fig. 5). Good clinical evolution was seen.

This case illustrates the added value of cholangioscopy in the diagnosis of intraductal papillary neoplasm of the bile duct.
duct, both for anatomical localization (and exclusion of lesion in the left intrahepatic ductal system) as well as for histopathological confirmation by intraductal biopsies. We believe cholangioscopy should be considered in every probable case of intraductal papillary neoplasm of the bile duct.

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Competing interests

The authors declare that they have no conflict of interest.

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References


Fig. 4 Biopsy specimens confirmed the diagnosis of multifocal intraductal papillary neoplasm of the bile duct with low-grade intraepithelial neoplasia.

Fig. 5 Pathologic examination of the right hepatectomy specimen shows intraductal papillary neoplasm of the bile duct with intestinal type differentiation (MUC1 negative, diffuse MUC2 positive, focal MUC5AC positive, limited MUC6 positive).

Bibliography

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