



Photodynamic therapy for hepatic hilar intraductal papillary neoplasm of the bile duct: a case report

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INTRODUCTION

Intraductal papillary neoplasm of the bile duct (IPNB) is a rare disease that is characterized by papillary or villous neoplasm within the lumen of the bile duct.¹ IPNB can develop anywhere along the biliary tree.² In principle, aggressive resection is recommended as the first choice. However, hepatectomy is too aggressive for small localized tumors without significant invasion. Here, we report a case of a small hilar IPNB that was diagnosed using a digital cholangioscope (SpyGlass, Boston Scientific, Marlborough, Mass) and treated by cholangioscopy-guided photodynamic therapy (PDT) (Guoyi Huake, Suzhou, China).

CASE REPORT

A 54-year-old man was admitted to our department with recurrent cholangitis. CT and MRCP revealed a cystic dilation of the hepatic hilar and right intrahepatic bile ducts (Fig. 1). The levels of tumor markers, such as alkaline phosphatase, carcinoembryonic antigen, and carbohydrate antigen 19-9, were normal.

ERCP was performed by using a nonradiation strategy. Successful biliary cannulation was confirmed by visible bile aspiration and biliary view (Fig. 2).³

The cholangioscope was advanced into the bile duct for a biliary view, and a large amount of mucin was found, which was removed by repeated basket extraction and gentle cholangioscopy-guided water irrigation. Lateral flat lesions with fish-egg-like papillary projections were found at the orifice of the right intrahepatic bile duct. Targeted biopsies by SpyBite were performed, and histopathological examination showed IPNB with low-grade intraepithelial neoplasia (Fig. 3).⁴

Because surgical resection is traumatic, the patient repeatedly refused surgery. For a further treatment plan, we attempted resection via cholangioscopy-guided PDT (Guoyi Huake) because the size of the lesions was small without obvious signs of invasion at the preoperative examination (Video 1, available online at www.giejournal.org).

A PDT optical fiber was introduced near to the lesions (excitation light at 630-690 nm was delivered for 20 minutes at a dose of 250 J/cm²) and was ceased when a pale view of the lesions was present (Fig. 4). A 7F plastic biliary stent (Mircotech, Nanjing, China) was inserted after the intervention. No adverse events occurred during or after the procedure. Follow-up magnetic resonance imaging (after 1 month) revealed less cystic dilation of the right intrahepatic bile duct than before the procedure and a slight dilation of the extrahepatic bile duct (Fig. 5). Follow-up cholangioscopy (after 4

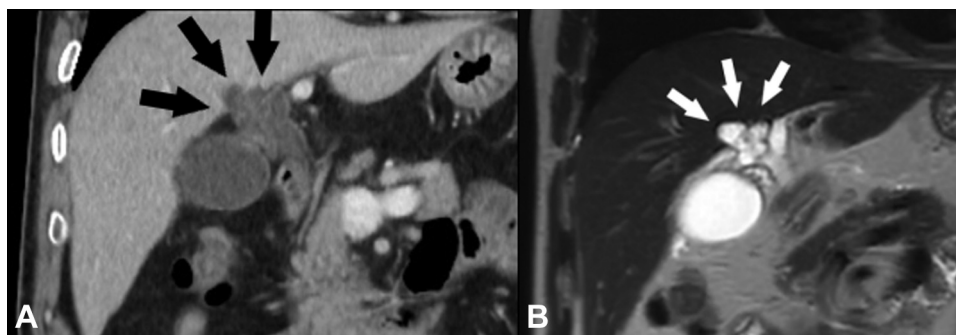


Figure 1. **A**, CT showing cystic dilation of the hilar bile ducts (*black arrows*). **B**, MRCP showing dilation of the hilar and right intrahepatic bile ducts (*white arrows*).

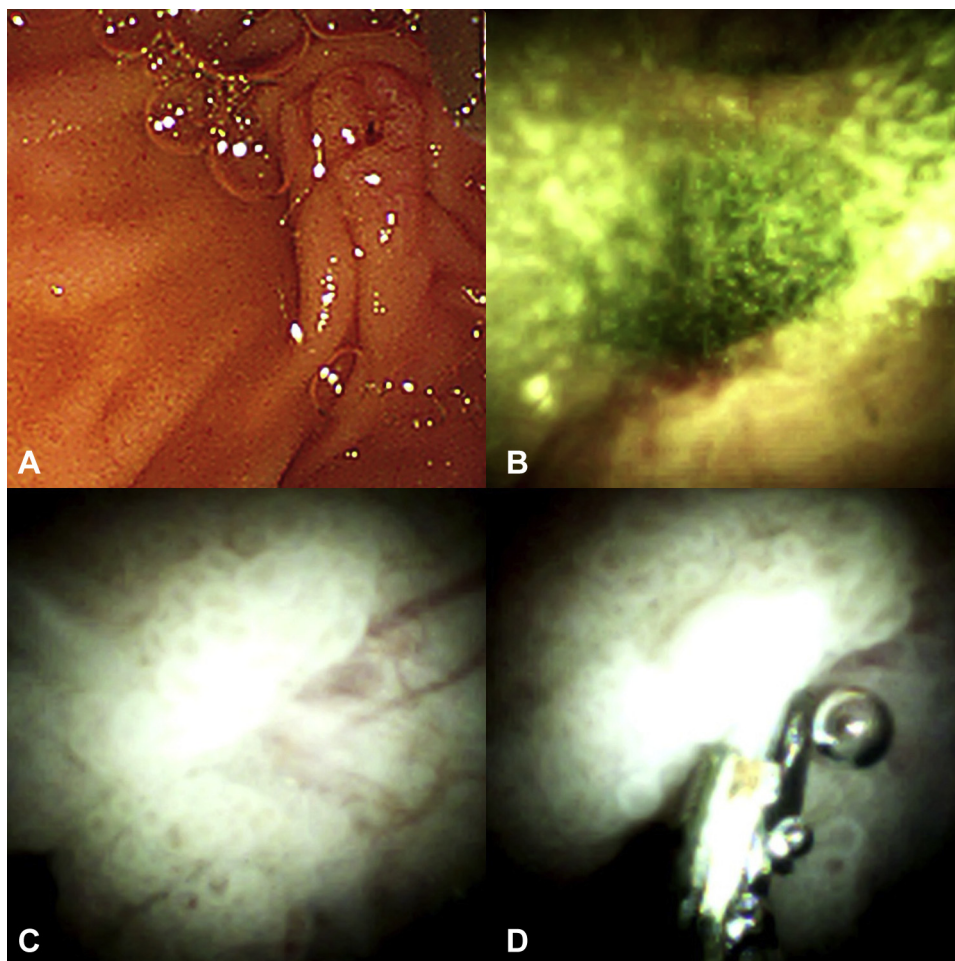


Figure 2. **A**, Fish-egg-like endoscopic appearance of the ampulla of Vater. **B**, Cholangioscopy showing a significant amount of intraductal mucin. **C**, Cholangioscopy showing multiple small areas of fish-egg-like papillary projections at the orifice of the right intrahepatic bile duct. **D**, Biopsy using SpyBite forceps under direct visualization.

months) showed fibrosis scars, and no lesion residual was present (Fig. 6).

DISCUSSION

Conventional ERCP is not technically sufficient for the diagnosis of local dilation of the biliary duct. Novel digital cholangioscopy provides an ideal tool for such lesions. Because of the advantages of the novel cholangioscopy,⁵ the diagnosis of this local IPNB was established by direct visualization and pathological outcome. Digital cholangioscopy also provides a novel platform for precise intervention for biliary malignancies. Saumoy et al⁶ have reported their experience with cholangioscopy-targeted PDT for unresectable cholangiocarcinoma. Considering the enormous damage caused by

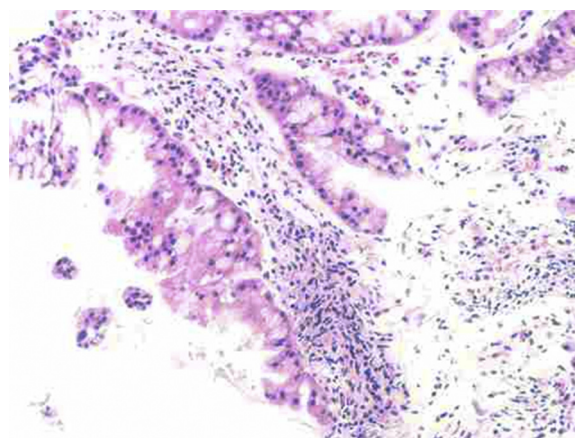


Figure 3. Histopathological discovery of atypical cells, papillary and epithelial configuration of the bile duct with mucinous metaplasia, and focal epithelium with low-grade dysplasia (H&E, orig. mag. $\times 40$).

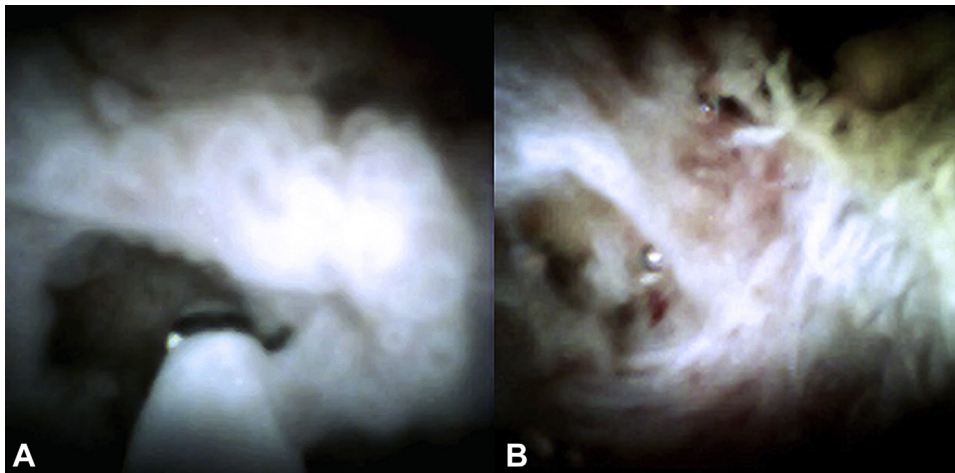


Figure 4. **A,** A photodynamic therapy optical fiber was introduced near the lesions by cholangioscopy. **B,** Cholangioscopy showing inflammatory changes, tumor necrosis, and exfoliated tissues in the bile duct after photodynamic therapy.

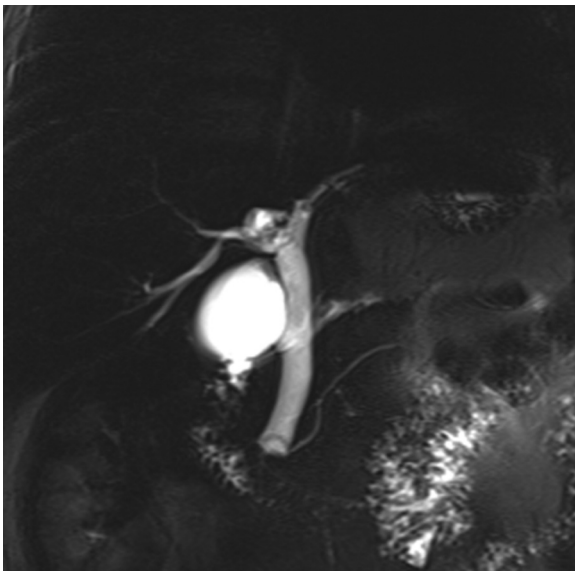


Figure 5. Followed-up MRCP showed less cystic dilation of the right intrahepatic bile duct than before the treatment and a slight dilation of the extrahepatic bile duct.

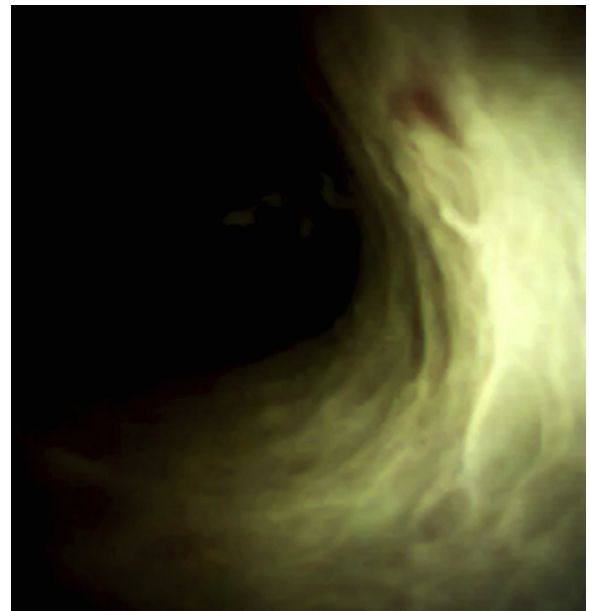


Figure 6. Fibrosis scars were visible at the site of the initial damage, and biliary inflammation disappeared with smooth duct walls.

hepatectomy, treatment methods that preserve organ function are worth trying. Two types of destructive platforms, PDT and radiofrequency ablation (RFA),⁷ are available for biliary neoplasm.

Currently, cholangioscopy-guided RFA is not available. This is because the working channel does not permit the passage of the probe of RFA. However, because of the small diameter (0.9 mm) of the PDT radiation probe, it can be introduced into the working channel smoothly. Hence, an intraluminal, direct-visualization-guided PDT is possible, and a targeted intervention is technically possible. Our intervention was based on the direct visuali-

zation of cholangioscopy and is similar to the previous report by Saumoy et al.⁶

Compared with surgical resection, this cholangioscopy-guided therapy is advantageous in achieving precise treatment for such circumscribed lesions. Based on follow-up imaging and endoscopy, this intervention demonstrated a novel therapeutic outcome.

DISCLOSURE

All authors disclosed no financial relationships.

Abbreviations: IPNB, intraductal papillary neoplasm of the bile duct; PDT, photodynamic therapy; RFA, radiofrequency ablation.

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