Recurrent extrahepatic cholangiocarcinoma after bile duct resection for intraductal papillary mucinous neoplasm of the bile duct

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Intraductal papillary neoplasm of the bile duct (IPNB) is a rare but recognized precursor to invasive carcinoma of the biliary tree and is characterized by dilated intrahepatic ducts with mucinous or papillary epithelium. Progression of papillary biliary neoplasia from benign disease to malignancy is thought to be similar to the pathway of an intraductal papillary neoplasm of the pancreas. The mainstay of therapy has been surgical resection. Surgical series demonstrate high rates of invasive cancer arising from IPNB; rates range from 40% to 70%. Very little is known about the prognostic variables in papillary bile duct tumors.

CASE REPORT

A 54-year-old man had presented 2 years earlier with fatigue, abdominal pain, and jaundice. Magnetic resonance imaging (MRI) and MRCP showed evidence of a bile duct mass extending to the margin of the intrapancreatic portion of the distal common bile duct. The bile duct was dilated to 2.4 cm (Fig. 1). ERCP demonstrated excessive mucin in the bile duct, and brushings were consistent with adenocarcinoma. He subsequently underwent excision of the extrahepatic bile duct and Roux-en-Y hepaticojejunostomy to the intrahepatic bile ducts. Notably, the bile duct was excised to the point where it tapered to a normal caliber.

A frozen section intraoperatively did not show dysplasia or malignancy at the resection margins; therefore, pancreaticoduodenectomy was not performed. Pathologic analysis demonstrated a focus of well-differentiated adenocarcinoma arising from an IPNB. Surgical margins and resected lymph nodes were negative. Surveillance MRI was performed every 6 months. Two years after the initial resection, imaging showed a new expansile hypoenhancing soft tissue lesion in the intrapancreatic portion of the remnant bile duct (Fig. 2; Video 1, available online at www.VideoGIE.org). An ERCP was performed for further evaluation and tissue sampling.

PROCEDURE

An ERCP was performed through the native ampulla, and a wire or other instruments could not be passed beyond 3 cm proximally, likely because of the known extrahepatic bile duct resection. Debris and mucin emanated from the biliary orifice (Fig. 3). Direct cholangioscopy was performed for improved visualization and demonstrated

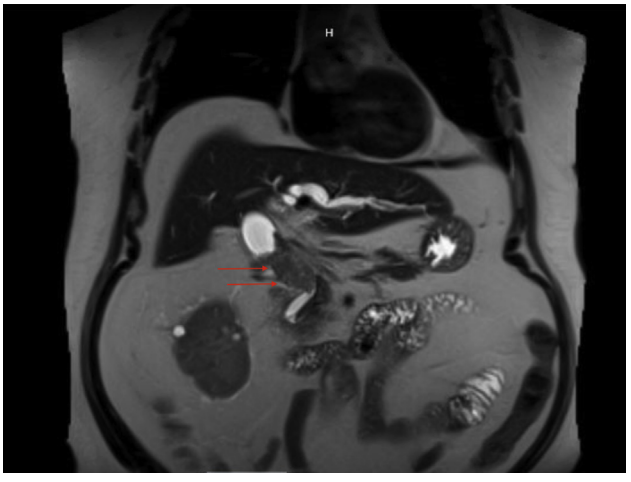

Figure 1. Initial magnetic resonance image from 2 years prior depicting lesion in bile duct.

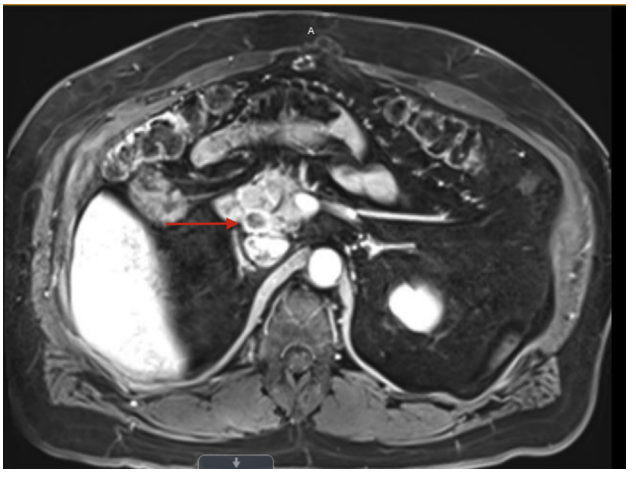

Figure 2. Surveillance magnetic resonance image demonstrating recurrence of intraductal papillary neoplasm from the remnant bile duct.

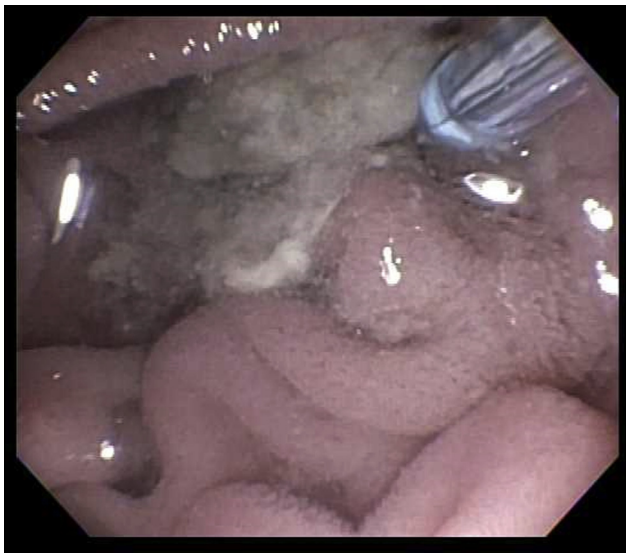

Figure 3. Extrusion of mucin and debris with cannulation.

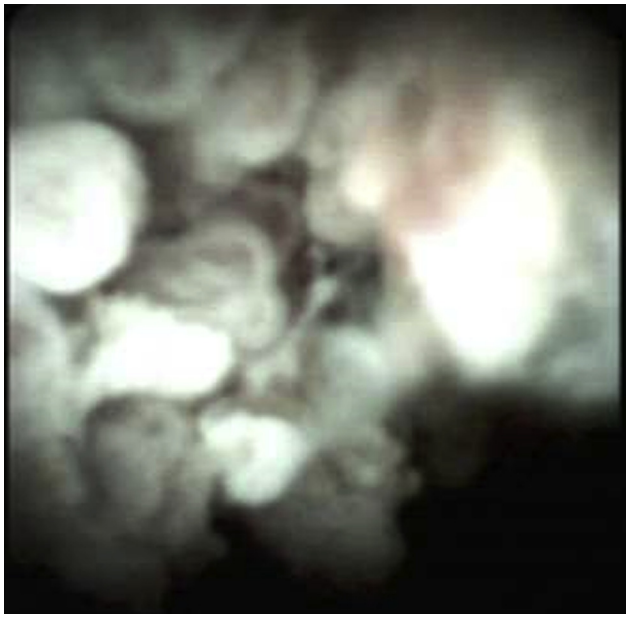

Figure 4. Direct cholangioscopic view demonstrating recurrent tumor in the bile duct.

frondy tissue growth in the extrahepatic bile duct concerning for recurrence (Fig. 4). Biopsy specimens were taken from this tissue (Fig. 5).

OUTCOME

Examination of the biopsy specimens confirmed recurrent adenocarcinoma arising from an IPNB. The patient underwent uncomplicated pancreaticoduodenectomy for definitive management (Fig. 6). Investigation of the

Figure 5. Fluoroscopic depiction of biopsy of the lesion, showing length of the residual bile duct.

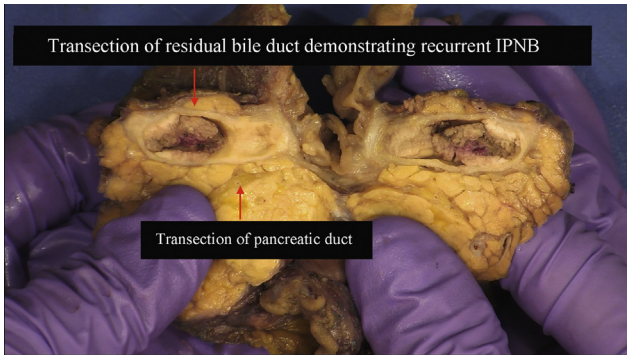

Figure 6. Gross specimen after pancreaticoduodenectomy.

hepaticojejunostomy by device-assisted enteroscopy may be considered in the future because IPNB can be multifocal.

CONCLUSION

Surveillance after resection of IPNB is critical in patients with residual extrahepatic bile duct, even in patients with negative resection margins. Aggressive further resection should be considered when the resection margin is involved by any residual lesion including dysplasia in IPNB. Direct cholangioscopy should be considered as an adjunctive therapy to facilitate direct visualization and

diagnostic sampling, especially in cases where advancement of the wire deep in the remnant bile duct is not feasible.

DISCLOSURE

Dr Schulman is a consultant for Apollo Endosurgery, Boston Scientific, and Microtech. All other authors disclosed no financial relationships relevant to this publication.

Abbreviations: IPNB, intraductal papillary neoplasm of the bile duct; MRI, magnetic resonance imaging.

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