Recurrent cholangitis due to intraductal papillary mucinous neoplasm of a cystic duct remnant

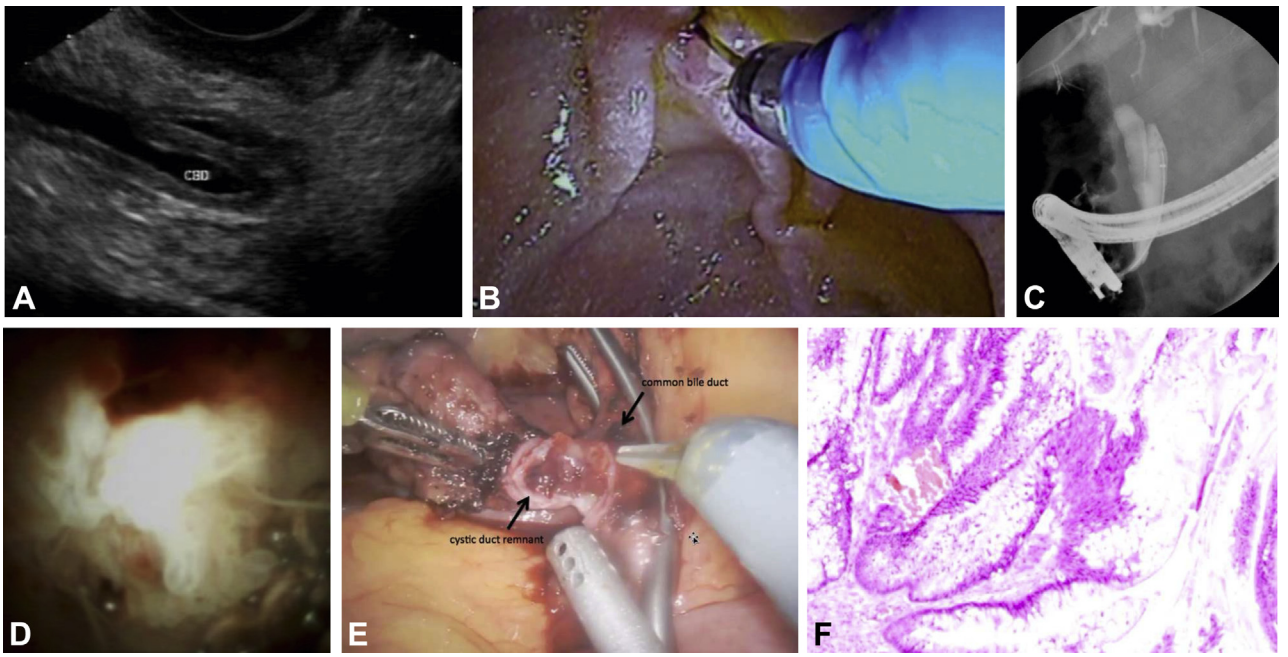

Figure 1. A, EUS image of a thickened common bile duct (CBD) with hypoechoic wall thickening. **B,** Endoscopic view of cholangioscope and guidewire cannulation of bile duct. **C,** Fluoroscopic view of cholangioscope in cystic duct. **D,** Cholangioscopic view of cystic duct remnant showing a villous tumor with numerous frondlike projections. **E,** Intraoperative view of laparoscopic robotic excision of the cystic duct with primary closure of the common bile duct. **F,** Histologic view of cystic duct remnant showing intraductal papillary mucinous neoplasm with low-grade to intermediate-grade intraepithelial dysplasia (H&E, orig. mag. × 40).

A 71-year-old man presented with 6 months of intermittent, sharp, right-sided abdominal pain, fevers, jaundice, and a 15-pound weight loss. He had undergone cholecystectomy 12 years earlier for symptomatic cholelithiasis. The initial presentation was thought to be due to biliary pancreatitis; ERCP at the referring institution revealed a normal bile duct with no filling defects. Sphincterotomy was performed, and it was assumed that he had passed a stone. His symptoms recurred, and 2 months later he underwent repeated ERCP. It demonstrated drainage of mucinous debris from the duct during a balloon sweep. EUS showed a thickened bile duct and sludge in the proximal cystic duct remnant (Fig. 1A). Test results for immunoglobulin-G4 and antinuclear antibody were negative. Two months later, he experienced recurrent symptoms consistent with biliary obstruction and underwent a third ERCP. Again, a large mucinous plug was removed from the bile duct. He was then referred to our center for further evaluation.

We performed ERCP with cholangioscopy using the Spyglass system (Boston Scientific, Marlborough, Mass) (Video 1, available online at www.VideoGIE.org). The main papilla was notable for a widely patent biliary orifice consistent with prior sphincterotomy but without obvious mucinous drainage. The bile duct was cannulated by use of a wire-guided technique (Fig. 1B), and fluoroscopy was used to confirm the position of the cholangioscope in the cystic duct remnant (Fig. 1C). Cholangioscopic views of the entire bile duct and cystic duct orifice were normal. After the cystic duct remnant was entered, cholangioscopy revealed a villous tumor 4 centimeters proximal to the hepatocystic duct junction and abundant mucus (Fig. 1D). The tumor had numerous frondlike papillary projections, and intraductal biopsy specimens were taken. Pathologic examination showed fragments of a tubulovillous adenoma.

Because of a high suspicion for biliary intraductal papillary mucinous neoplasm (IPMN), the patient was referred to surgery. He underwent laparoscopic robotic excision of the cystic duct and primary closure of the common

Written transcript of the video audio is available online at www.VideoGIE.org.

bile duct (Fig. 1E). Pathologic examination showed a 1.3-centimeter IPMN of the cystic duct with low-grade to intermediate-grade intraepithelial dysplasia and negative margins on resection (Fig. 1F). IPMN of the biliary tract bears a striking resemblance to the more commonly recognized IPMN of the pancreas and should be considered in the differential diagnosis of patients with recurrent cholangitis and pancreatitis.

DISCLOSURE

Dr Mejia is a surgeon with Intuitive. Dr Kedia and Dr Tarnasky are consultants and speakers for Boston Scientific. All other authors disclosed no financial relationships relevant to this publication.

Umangi Patel, MD, Alejandro Mejia, MD, *Methodist Dallas Medical Center,* **William Torres, MD,** *East Texas Gastroenterology, Longview, Tex,* **Prashant Kedia, MD, Paul Tarnasky, MD,** *Methodist Dallas Medical Center, Dallas, Texas, USA*

http://dx.doi.org/10.1016/j.vgie.2017.08.014