



Single-operator cholangioscopy monitoring of a remaining bile duct after congenital choledochal cyst surgery: a case report with an innovative approach

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INTRODUCTION

Congenital choledochal cysts are rare and generally associated with an anomalous pancreaticobiliary ductal union (APBDU).¹ Diagnosis can be done before birth and most often is done during childhood. Symptoms include abdominal pain, nausea, vomiting, and jaundice.² APBDU is characterized by an abnormal junction between the lower choledochus and the terminal part of the pancreatic duct. APBDUs are most often due to a fusion anomaly between the bile duct and the pancreatic duct during embryogenesis. Three anatomic criteria have been proposed to define APBDU: a common duct longer than 15 mm, an extra duodenal biliopancreatic junction at a distance from the sphincters, and a connection angle of over 30 degrees.

The risks associated with APBDU are lithiasis and biliary tract cancers such as cholangiocarcinoma or gallbladder cancer. These are due to pancreatic-biliary reflux with pancreatic juice stasis in the bile duct.³ Treatment consists of as complete a resection as possible of the cystic choledochal dilatation and confection of a biliary-digestive anastomosis.⁴ Because the distal part of the cyst is often intrapancreatic or retropancreatic, resection sometimes leaves a small biliary cyst remnant that remains exposed to corrosive pancreatic reflux. Some surveillance of that remnant is therefore generally advocated during the lifetime owing to the risk of malignant transformation. We present a case of Spyglass choledochoscope (Boston Scientific Corporation, Natick, Mass, USA) monitoring of a remaining bile duct after congenital choledochal cyst surgery.

CASE PRESENTATION

A 29-year-old woman presented to our clinic with right upper abdominal pain. She had undergone surgery as a child for a congenital choledochal cyst and had a hepaticojejunostomy. Hepatic tests were normal, and C-reactive protein was negative. On ultrasound, there was doubt about the presence of gallstones in the bile duct. Magnetic resonance imaging confirmed the presence of stones in the lower part of the main bile duct, which was dilated up to 13 mm in diameter (Fig. 1); the main pancreatic

duct was measured at 3 mm above the intraductal stones. EUS confirmed these findings, including the presence of a 15-mm stone in the bile duct.

An ERCP confirmed the dilation of the remaining bile duct, with filling defects on contrast injection (Fig. 2). A 15-mm stone was removed after ampullary dilation and sphincterotomy. Subsequently, pancreatic cannulation showed another filling defect in the pancreatic head. After hydrostatic dilation, a pancreatic prosthesis was placed (Zimmon, 7F, 12 cm, Cook Medical, Winston-Salem, NC).

Two months later, a second ERCP was performed. The pancreatic prosthesis was removed. There were no fluoroscopic defects in the bile duct. The remaining bile duct was measured at 12 mm in width and 20 mm in height. The narrow junction between the remaining part of the bile duct cyst and the common biliopancreatic duct was 3 mm long. The APBDU appeared to belong to Todani type I (fusiform) and to the P-C type according to Kimura,⁵ although depiction was uncertain because of ductal changes caused by the long-standing presence of stones (Fig. 3). We extracted the 2 remaining pancreatic stones with an extraction balloon before inserting a choledochoscope (10F outer diameter, 230 cm length) for exploration of the remaining bile duct. The mucosa was regular, monochromatic, and with no suspicious areas. We performed 5 biopsies with a Spybite miniforceps (Boston Scientific Corporation, Natick, Mass): 3 in the remnant cyst and 2 in the junction between the cyst and the choledochus (Video 1, available online at www.giejournal.org). Pathological results showed mucosal remodeling in the form of scarring without any malignancy. After an uneventful recovery and before discharge, the patient was offered a 3-year interval for a surveillance choledochoscope procedure to be planned.

DISCUSSION

This is, to our knowledge, the first case describing choledochoscope monitoring of a choledochal cyst remnant after childhood surgery for a type I cyst associated with APBDU. The incidence of malignant transformation of the bile duct after the age of 20 has been estimated at 14% and over 50% at 50 and beyond.⁶ Long-term follow-up

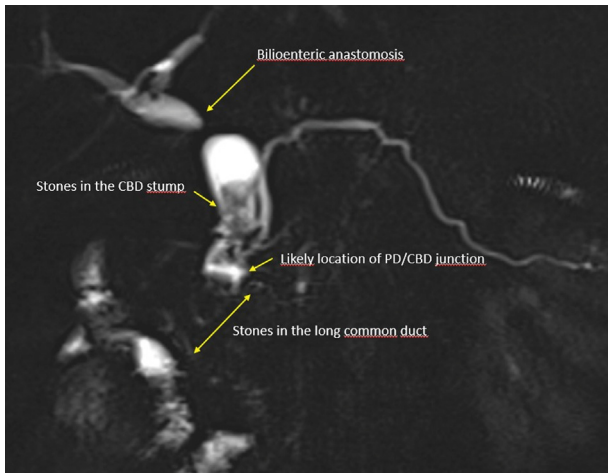


Figure 1. Biliary magnetic resonance imaging showing stones in the lower part of the main bile duct. *CBD*, Common bile duct; *PD*, pancreatic duct.

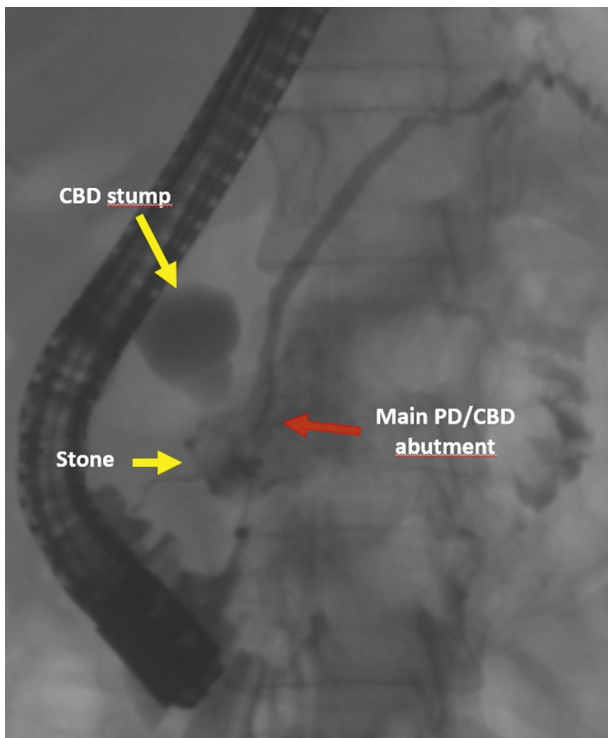


Figure 2. Fluoroscopic image of the first ERCP showing dilation of the remaining bile duct with filling defects on contrast injection. *CBD*, Common bile duct; *PD*, pancreatic duct.

is more than necessary in those patients. CT- or magnetic resonance imaging–based imaging is unable to disclose discrete changes in the thickness of the remaining bile duct and cyst remnant. EUS, although having a higher spatial resolution, is not accurate enough and cannot detect dysplasia, nor can biliary brushings during ERCP, which cannot be precisely targeted and are known to have a low yield. Retrograde single-operator cholangio-

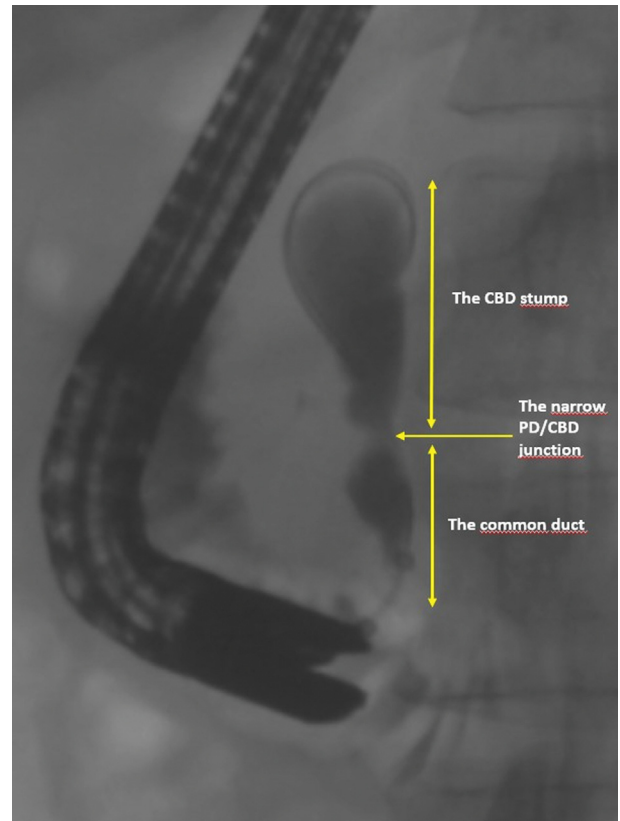


Figure 3. Fluoroscopic image of the second ERCP after extraction of the 2 remaining pancreatic stones. *CBD*, Common bile duct; *PD*, pancreatic duct.

scopy has the advantage of allowing a high-quality visual analysis of the mucosa, including surface and color changes as well as vascular anomalies and the ability to target biopsies. A limitation is the requirement for a sufficiently large outlet to allow the passage of the 10F catheter, which may need prior hydrostatic dilation, with an associated risk of pancreatitis. This was not required in our patient, whose case was also peculiar for the uncommon presence of pancreatic stones in the biliary remnant and the common duct.

CONCLUSIONS

Choledochoscope monitoring is suggested as an interesting alternative for surveillance of the remnant bile duct after choledochal congenital cyst surgery.

DISCLOSURE

Dr Prat is a consultant for Olympus Europe and Boston Scientific. All other authors disclosed no financial relationships.

Abbreviation: APBDU, anomalous pancreaticobiliary ductal union.

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