

## Dysplastic progression of a choledochal cyst on video cholangioscopy

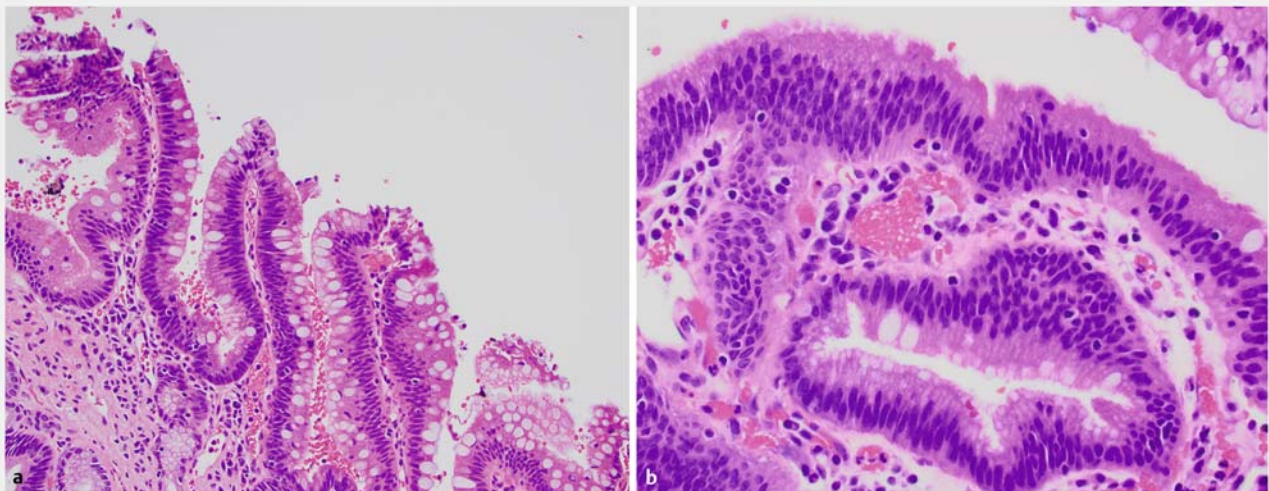
Choledochal cysts are rare congenital cystic dilatations of the biliary tree. Surgical resection is felt to reduce the risk of malignant degeneration, which can be seen in 40%–60% by the fifth decade [1,2]. Proposed mechanisms for malignancy include reflux of pancreatic juices in the setting of an anomalous pancreaticobiliary junction (APBJ) causing chronic inflammation or bile stasis, leading to “carcinogenic” bile acid generation [3]. We present a patient with choledochal cysts where serial cholangioscopy demonstrated visual and pathologic evidence of the metaplasia to dysplasia sequence. The patient was a 47-year-old woman who had previously undergone cholecystectomy and presented with ongoing right upper quadrant pain and a dilated common bile duct on imaging, along with mild elevations in alkaline phosphatase and transaminases. Endoscopic retrograde cholangiopancreatography (ERCP) revealed retained stones and a cystically dilated distal bile duct suspi-

cious for a type III choledochal cyst (choledochocele). The cyst and long common channel were consistent with APBJ and biopsies revealed intestinal

metaplasia. The patient opted to undergo annual surveillance with alternating magnetic resonance cholangiopancreatography (MRCP) and cholangioscopy.



**Video 1** Video cholangioscopy and confocal endomicroscopy of a choledochal cyst in a patient undergoing annual surveillance.



**Fig. 1** Histopathological images (hematoxylin and eosin [H&E] stained) showing: **a** a representative section of the choledochal cyst in the resected specimen (magnification  $\times 200$ ) with villiform mucosa lined by epithelium with a predominantly intestinal phenotype and globular mucin droplets within many of the epithelial cells that are diagnostic of intestinal metaplasia; **b** a focus of low grade dysplasia arising from a background of intestinal metaplasia within the choledochal cyst in the resected specimen (magnification  $\times 400$ ), which is characterized by the loss of globular mucin droplets within the epithelium, in conjunction with hyperchromatic, elongated, pseudostratified epithelial cell nuclei. Source: Dr. Jeffrey Kaplan, University of Colorado.

The index video cholangioscopy (Olympus XCHF-B140Y, Tokyo, Japan) showed intestinal mucosa within the cyst and pathology showed intestinal metaplasia. Repeat video cholangioscopy 2 years later (Olympus TJF-Y008/CHF 280K003) revealed “tumor vessels” and probe-based confocal endomicroscopy (Cellvizio, Mauna Kea, Paris, France) demonstrated three malignant Miami criteria, including thick dark bands, dark clumps, and epithelial structures concerning for malignancy [4] (► **Video 1**). Biopsies demonstrated atypia but, because of the advanced imaging findings, the patient was referred for pancreaticoduodenectomy. The resected specimen revealed low grade dysplasia (► **Fig. 1**).

This case depicts the progression of a choledochal cyst from intestinal metaplasia to dysplasia documented by video cholangioscopy and provides further support for the aforementioned sequence of carcinogenesis in choledochal cysts. We also demonstrate suspected dysplasia by video cholangioscopy and confocal endomicroscopy that aided the decision-making process to proceed with surgery, despite non-confirmatory sampling for neoplasia.

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## Competing interests

Raj J. Shah is a consultant and advisory board member for Boston Scientific and a consultant for Olympus and Cook Medical. The remaining authors declare that they have no conflict of interest.

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