

Biliary papillomatosis diagnosed with mother-daughter narrow-band imaging (NBI) cholangioscopy



Fig. 1 Cholangiographic image demonstrating a dilated bile duct containing amorphous filling defects consistent with mucus plugs of biliary papillomatosis.

A previously healthy 47-year-old man presented to our institution with a 6-month history of recurrent episodes of jaundice and pruritus. Over this period a number of endoscopic retrograde cholangiographies (ERCs) had been undertaken, demonstrating mucin at the ampullary orifice and a dilated biliary system packed with mucin plugs (▶ **Fig. 1**).

Repeated biliary stenting had been performed for symptomatic relief. On referral to our institution, the patient was jaundiced with a bilirubin of 426 $\mu\text{mol/L}$ (normal range 5.1 – 17.0 $\mu\text{mol/L}$).

A mother-daughter video cholangioscopy with narrow-band imaging (NBI) capability (CHF-Y0002, Olympus, Melville, New York, USA) was carried out on the suspicion of presence of a mucin-secreting tumor within the biliary tree. A nasobiliary drain was placed for 48 hours to facilitate irrigation of the biliary tree. At cholangioscopy, multiple lesions, characterized by a fish egg-like appearance, were identified at

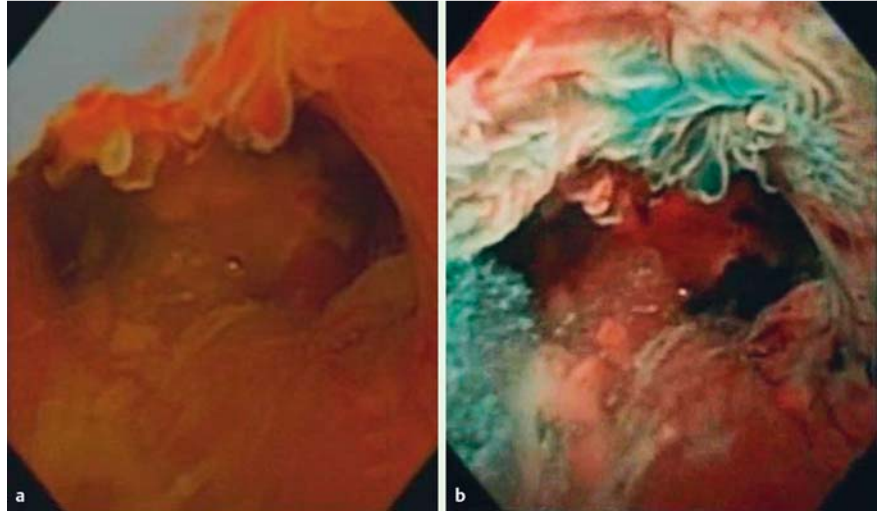


Fig. 2 Cholangioscopic image demonstrating the characteristic fish egg-like appearance of biliary papillomatosis using: **a** white light and **b** narrow-band imaging (NBI).

the bifurcation of the intrahepatic ducts and in the distal bile duct just proximal to the ampulla, consistent with multicentric papillomatosis (▶ **Fig. 2**, ▶ **Video 1**).

The patient was subsequently referred for consideration for liver transplantation and Whipple resection. However, in the course of this workup, he was deemed unsuitable for surgical resection because of the spread of tumor to surrounding lymph nodes. The patient has since been managed endoscopically with bilateral hilar stenting without complication.

Biliary papillomatosis, first reported by Chappet in 1894, is a rare disease characterized by multiple papillary adenomas in the biliary tree [1]. It should be considered a premalignant condition with a high malignant potential [2]. The disease is characterized by relapsing episodes of acute cholangitis and obstructive jaundice resulting from intermittent biliary obstruction from secreted mucin and the tumor itself, and secondary cirrhosis is often present by the time of presentation [3]. ERC has a key role in the diagnosis of biliary papillomatosis. Diffuse bile duct dilatation associated with amorphous filling defects are characteristic findings at cholangiography. However, a large amount of mucin secretion and obstruction by the tumor prevent complete opacification of the entire biliary tract. As a result, precise

evaluation of ductal extent by ERC is often suboptimal. Diagnosis is usually confirmed by tissue biopsy either through peroral or percutaneous transhepatic cholangioscopy [2].

Peroral cholangioscopy facilitates noninvasive visualization of the biliary system. While the conventional method involved the use of fiberoptic mother-daughter cholangioscopy, its use is becoming less common with the advent of the Spyglass system. However, our case, in addition to the report by Itoi et al., demonstrates that NBI video cholangioscopy provides superior images in terms of quality and size in contrast with both Spyglass and fiberoptic cholangioscopy [4, 5]. Video cholangioscopy in our patient detailed clearly the multicentric extent of the disease, thereby having the potential to guide the appropriate surgical procedure. In addition, mucosal imaging techniques such as NBI may facilitate improved characterization and targeted biopsies of abnormal lesions in the biliary tract similar to their emerging role in the gastrointestinal tract [6].

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Competing interests: None

Video 1

Biliary papillomatosis diagnosed with mother-daughter narrow-band imaging (NBI) cholangioscopy. The cholangioscope is positioned in the common bile duct. Normal biliary epithelium is seen first with white light and subsequently with NBI. Mucin and the fish egg appearance of the papillary biliary tumor is demonstrated with both white light and NBI.

**F. Donnellan, M. P. Swan, G. R. May,
P. P. Kortan**

Center for Therapeutic Endoscopy and
Endoscopic Oncology, St Michael's
Hospital, Toronto, Ontario, Canada

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Corresponding author

Dr. F. Donnellan

Center for Therapeutic Endoscopy and Endoscopic
Oncology

St. Michael's Hospital

Toronto

Ontario

Canada

fdonnellan77@hotmail.com