Diagnosis of biliary cystadenoma by peroral video cholangioscopy

A 31-year-old woman with jaundice was admitted to our hospital. Computed tomography, magnetic resonance cholangiopancreatography, and endoscopic retrograde cholangiography suggested the presence of a solitary cystic lesion in the common bile duct (CBD) (Fig. 1 a, b, c).

Subsequent endoscopic ultrasonography (EUS) and intraductal ultrasonography (IDUS) showed a multilocular cystic tumor with cyst-in-cyst pattern in the CBD (Fig. 1 d; Fig. 2).

After the patient had recovered from jaundice, peroral video cholangioscopy (POCS) was performed. POCS revealed that the tumor had a flat surface with a fine network of thin vessels, suggesting that it was a benign tumor (Fig. 3). Based on these findings, a preoperative diagnosis of biliary cystadenoma (BCA) was made. The patient underwent choledochal resection and subsequent choledochojunostomy. The resected specimen included a 45-mm multilocular cystic mass in the CBD. Microscopically, the cyst wall was lined by a single layer of high columnar non-neoplastic epithelium, with underlying ovarian-like stroma. Spindle cells in the ovarian-like stroma were positive for progesterone and estrogen receptors.

BCAs are rare neoplasms that usually occur in the liver and, less frequently, in the extrahepatic bile ducts [1–4]. They are considered to originate from the embryonic tissue precursors of the biliary epithelium and to be premalignant lesions.

This is the first report of a diagnosis of extrabiliary cystadenoma by POCS prior to surgery. In this case, POCS showed that the tumor arose from the bile duct wall and had a smooth surface with a fine network of thin vessels, features that are compatible with a benign lesion [5] and are potentially characteristic findings of BCA. POCS clearly depicted the cystic tumor in the CBD. Imaging studies enabled preoperative diagnosis of BCA in the CBD. Such tumors are probably better termed ovarian tumor of the bile duct.

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Fig. 2  Intraductal ultrasonography: the multilocular lesion gradually compressed the wall of the common bile duct and finally occluded the lumen.

Fig. 3  Peroral cholangioscopy demonstrated a smooth and soft cystic lesion in the common bile duct.