A 57-year-old man was referred for a diagnostic workup because of elevated serum liver enzyme levels (γ-glutamyltransferase 53.94 μkat/L, reference range 0.00–0.63). A transabdominal ultrasound revealed dilated extrahepatic bile ducts, and on endoscopic ultrasound (EUS) the common bile duct (CBD) was dilated with hyperechoic foci with no acoustic shadowing within, and a 20-mm hyperechoic frond-like mass was noted within the infundibulum of gallbladder (● Fig. 1). The CBD and gallbladder walls were thin with smooth outer margins. Endoscopic retrograde cholangiography (ERC) revealed a 15-mm long filling defect within the dilated CBD (● Fig. 2). A biopsy sample taken from the CBD showed evidence of tubulopapillary adenoma with foci of well-differentiated adenocarcinoma. An abdominal computed tomography (CT) scan showed atypical, but noncontrast-enhancing, hypodense structures within the gallbladder infundibulum, which were not suggestive of tumor (● Fig. 3). The patient underwent open laparotomy, cholecystectomy, and extensive resection of the extrahepatic bile ducts with Roux-en-Y hepaticojejunostomy. The histological examination confirmed the diagnosis of papillomatosis of the extrahepatic bile ducts, with foci of high-grade dysplasia (● Fig. 4) and secondary growth extending into the gallbladder along the cystic duct.

Biliary papillomatosis (BP) is a rare disease characterized by the presence of numerous papillary adenomas within the intra- and/or extrahepatic biliary tree, with or without mucin hypersecretion [1]. It is a premalignant condition, undergoing malignant transformation in 25–50% of cases [1]. The diagnosis is based on histological examination of specimens obtained during ERC, cholangioscopy, or surgery. Papillary carcinoma, which manifests as a well-defined, polypoid mass obstructing the lumen of a single extrahepatic bile duct and as a single filling defect on ERC, should be distinguished from multicentric biliary papillomatosis.
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Fig. 4 The dilated common bile duct with papillary clusters of epithelial cells showing high-grade dysplasia (hematoxylin and eosin, magnification × 40).