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 tubulo-papillary adenoma with foci of well-differentiated adenocarcinoma. An abdominal computed tomography (CT) scan showed atypical, but noncontrast-enhancing, hyperdense 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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1 Department of Gastroenterology, Hospital Vitkovice, Ostrava, Czech Republic
2 Faculty of Medicine, University of Ostrava, Czech Republic
3 Department of Surgery, J. G. Mendel Oncology Centre, Novy Jicin, Czech Republic
4 Department of Pathology, P&R Laboratory, Novy Jicin, Czech Republic

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**Corresponding author**

M. Kliment
Department of Gastroenterology
Hospital Vitkovice
Zaluzanskeho 1192/15
703-84 Ostrava
Czech Republic
Fax: +420-595-633077
martin.kliment@nemvitkovice.cz

**Fig. 4** The dilated common bile duct with papillary clusters of epithelial cells showing high-grade dysplasia (hematoxylin and eosin, magnification ×40).