Adenomyomatous hyperplasia of the lower bile duct mimicking a papillary bile duct tumor

Fig. 1 Abdominal contrast-enhanced computed tomography (CT) scan showing a mass lesion (yellow arrows) in the lower bile duct: **a** in the axial plane; **b** in the coronal plane.

Adenomyomatous hyperplasia is an extremely rare benign lesion of the hepatobiliary tract, including the ampulla of Vater, with most cases described in the gallbladder [1–4]. However, its importance lies in its ability to mimic bile duct tumors [1,2]. A 66-year-old man without jaundice was found to have dilatation of the common and intrahepatic bile ducts on ultrasonography performed during ambulant treatment of diabetes mellitus and hypertension. An enhanced computed tomography (CT) scan showed a mass lesion in the lower bile duct (● **Fig. 1**). Endoscopic ultrasono-

graphy (EUS) revealed a hypoechoic lesion in the bile duct portion of the ampulla of Vater, with the lesion also extending to the lower bile duct (● **Fig. 2a**), suggesting a papillary bile duct tumor. EUS-guided fine needle aspiration (EUS-FNA) of the bile duct lesion was therefore performed, and microscopic examination showed no apparent malignant cells (● **Fig. 2b**).

Endoscopic retrograde cholangiography (ERCP) showed an irregular stricture in the lower bile duct (● **Fig. 3a**), while intraductal ultrasound and peroral cholangioscopy by SpyGlass demonstrated that this stricture was caused by a papillary mass (● **Fig. 3b,c**; ● **Video 1**). The endoscopic transpapillary biopsy specimen of this papillary mass showed no apparent malignant cells (● **Fig. 4**); however, the possibility of a tumor could not be completely excluded and we therefore performed pancreatoduodenectomy. Histologically, the bile duct epithelium was structurally papillary, and mildly atypical glands and hyperplasia of smooth muscle fibers were observed (● **Fig. 5**). The papillary bile duct lesion was finally diagnosed as adenomyomatous hyperplasia.

No specific imaging features permit the reliable differentiation of adenomyomatous hyperplasia from tumor; however, a possible diagnosis of adenomyomatous hyperplasia should be kept in mind in a patient with bile duct stricture. For accurate preoperative diagnosis, it is important to obtain enough biopsy specimens confirming hyperplastic glands surrounded by smooth muscle fibers.

Fig. 2 The lesion in the lower bile duct was shown: **a** on linear array endoscopic ultrasonography (EUS), to be a 14.9-mm hypoechoic mass (yellow arrows); **b** on histology from an EUS-guided fine needle aspiration (FNA), to have no apparent malignant cells (hematoxylin and eosin [H&E] stain, original magnification $\times 200$).

Video 1

Peroral cholangioscopy by SpyGlass showing that the stricture in the lower bile duct was caused by a papillary mass.

Fig. 3 Appearance of the lesion on: **a** endoscopic retrograde cholangiography showing an irregular stricture at the lower bile duct; **b** intraductal ultrasound; and **c** peroral cholangioscopy by SpyGlass showing that the stricture was caused by a papillary mass.

Fig. 4 Histological appearance of the endoscopic transpapillary biopsy taken from the papillary mass showed no apparent malignant cells (hematoxylin and eosin [H&E] stain, original magnification $\times 200$).

Fig. 5 Histological appearance of the hematoxylin and eosin (H&E)-stained specimen that was resected at pancreatoduodenectomy showed papillary hyperplasia of the surface mucosa and dilated glands surrounded by hyperplastic smooth muscle fibers at original magnifications of: **a** $\times 12.5$; **b** $\times 200$.

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Competing interests: None

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