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

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 ultrasonography (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.
Endoscopy_UCTN_Code_CCL_1AZ_2AC

Competing interests: None

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DOI http://dx.doi.org/10.1055/s-0042-115941
Endoscopy 2016; 48: E311 – E312
© Georg Thieme Verlag KG
Stuttgart · New York
ISSN 0013-726X

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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 × 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 × 12.5; b × 200.