Title/cover page

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Title: Adenomyomatosis hyperplasia arising in the bile duct

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A 46-year-old man was referred to our hospital for a detailed examination of the bile duct. Contrast-enhanced computed tomography showed enhanced wall thickness of the lower bile duct. Endoscopic ultrasonography (EUS) revealed a hypoechoic papillary lesion in the distal bile duct (Figure 1a), and endoscopic retrograde cholangiography showed a filling defect. Per-oral direct cholangioscopy (POCS) using an ultrathin endoscope (EG-L580NW; Fujifilm, Tokyo, Japan) revealed flat, elevated lesions with a slightly reddish color (Figure 1b). Papillary adenocarcinoma of the bile duct was suspected, and we performed a bile juice cytology and a direct biopsy using biopsy forceps. The pathological results showed no signs of malignancy, however he ultimately decided to undergo pancreaticoduodenectomy, as a definitive diagnosis of the bile lesion could not be obtained. The pathological findings revealed papillary hyperplastic lesions at the distal end of the bile duct, and bundles of smooth muscle cells were observed between the hyperplastic mucosa (Figure 2). The papillary lesion shows a biliary phenotype on CK19 staining (Figure S-1) and bundles of smooth muscle fibers were confirmed on Desmin staining (Figure S-2). Based on these findings, a final diagnosis of adenomyomatous hyperplasia was made.

Adenomyomatosis arising in the periampullary area is extremely rare and frequently mimics neoplasm of the bile duct. Detailed investigation through accumulation of cases is necessary in the future.

Conflict of interest

None declared.

Figure legends

Figure 1.

a: EUS shows a hypoechoic papillary lesion (arrow) in the distal bile duct. BD: bile duct

b: POCS shows flat, elevated lesions with a slightly reddish color at the end of the bile duct.

Figure 2.

a: Histological findings show hyperplastic papillary lesions at the distal end of the bile duct (H.E. staining)

b: Hyperplastic papillary lesions and smooth muscle cells can be seen between the hyperplastic mucosa (H.E. staining).

Supplemental figure

a: The papillary lesion shows a biliary phenotype on CK19 staining.

b: Bundles of smooth muscle fibers were confirmed on Desmin staining.