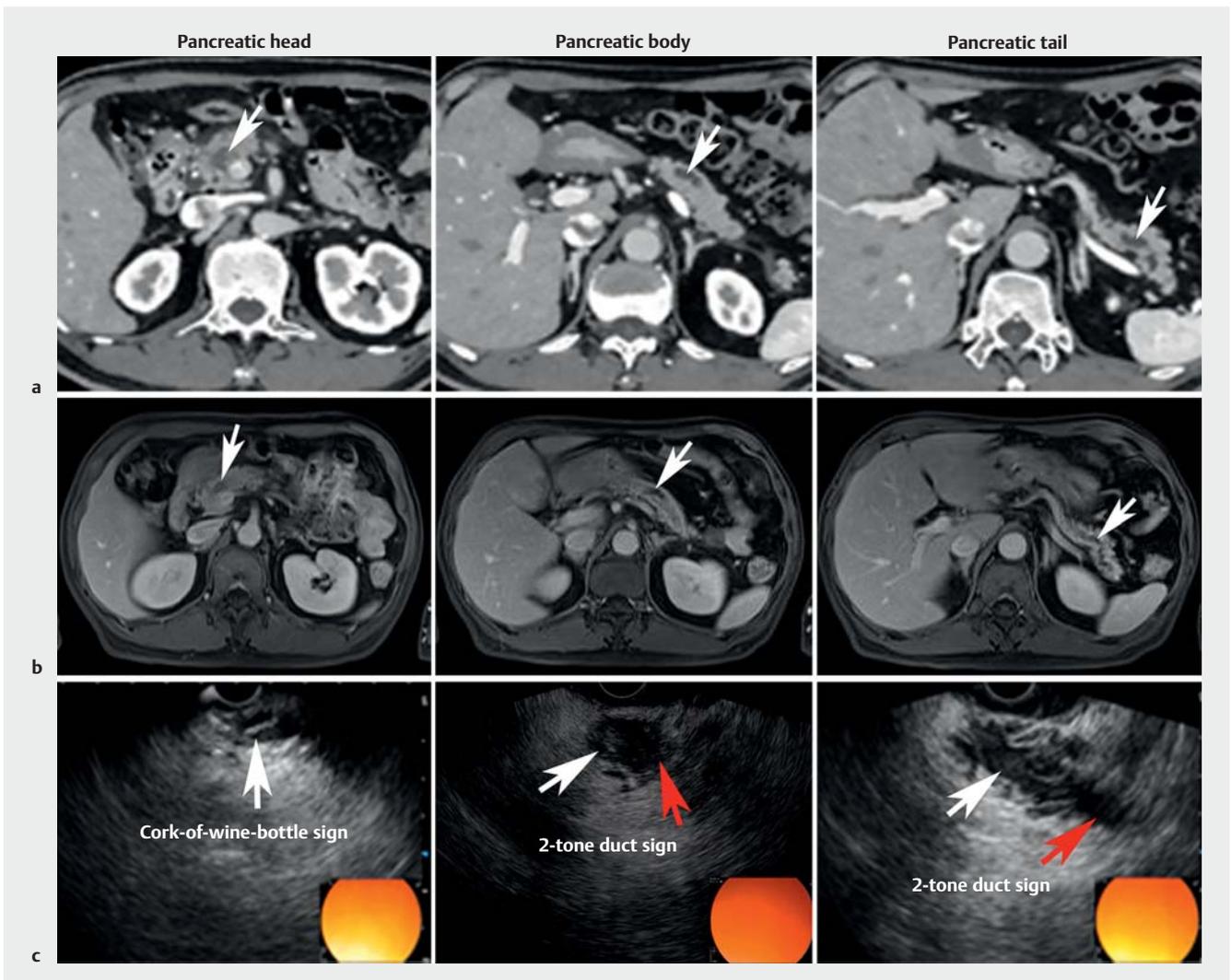


Preoperative diagnosis of multiple intraductal tubulopapillary neoplasms of the pancreas: dynamic imaging features from endoscopic ultrasound

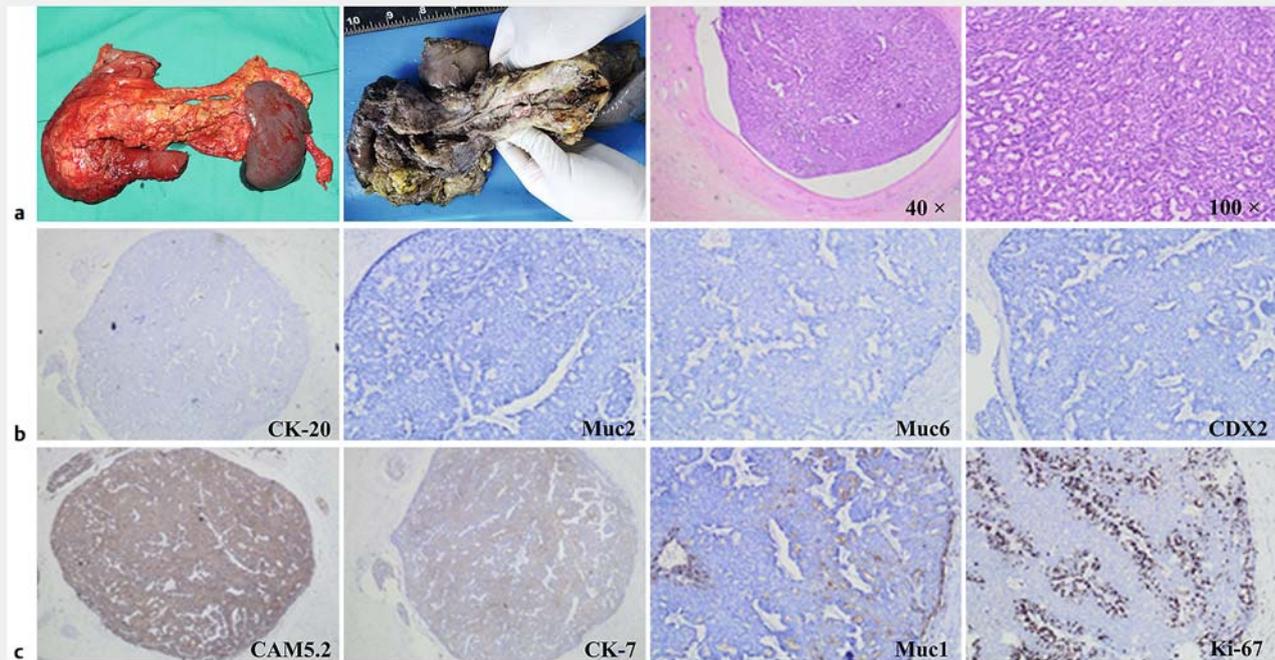
A 60-year-old man with recurrent pancreatitis was admitted to our hospital for further examination and treatment. Abdominal computed tomography highlighted multiple cystic lesions in the pancreas (► **Fig. 1 a**). Magnetic resonance imaging revealed atrophic pancreatic parenchyma and segmentally dilated

pancreatic duct (► **Fig. 1 b**). The diagnosis of intraductal papillary mucinous neoplasm (IPMN) or intraductal tubulopapillary neoplasm (ITPN) was proposed. Subsequent endoscopic ultrasound (EUS) showed multiple cystic lesions with solid tumors inside extending along the dilated main pancreatic duct (► **Fig. 1 c**). The

intraductal tumor was surrounded by pancreatic fluid in the head of the pancreas, representing the “cork-of-wine-bottle sign;” however, the hypoechoic tumors connecting with anechoic fluids were displayed as two different colors in the dilated duct, indicating the “2-tone duct sign” (► **Video 1**). Based on the



► **Fig. 1** Imaging manifestations of intraductal tubulopapillary neoplasm in a 60-year-old man. **a** Transverse dynamic computed tomography showed cystic lesions (arrows) in the dilated pancreatic duct. **b** Transverse magnetic resonance imaging also indicated multiple cystic lesions along the dilated duct in the head, body, and tail of the pancreas (arrows). **c** Transverse endoscopic ultrasound showed several intraductal tumors slightly protruding from the segmentally dilated pancreatic ducts, in which the “2-tone duct sign” (white and red arrows) and “cork-of-wine-bottle sign” (arrow) can be seen.



► **Fig. 2** Histopathologic characteristics of intraductal tubulopapillary neoplasm in a 60-year-old man. **a** Gross specimen presenting solid tumors without mucin located in the dilated pancreatic duct, and the histopathologic slides stained with hematoxylin and eosin showing the dilated main pancreatic duct containing a solid tumor with tubulopapillary structures. **b** Immunohistochemical staining revealed that the tumor was negative for CK-20, MUC2, MUC6, and CDX2. **c** Immunohistochemistry demonstrated positive staining for CAM5.2, CK-7, and MUC1. The proliferative index of Ki-67 reached 70%.



► **Video 1** Preoperative diagnosis of multiple intraductal tubulopapillary neoplasms of the pancreas based on the dynamic imaging features from endoscopic ultrasound, consisting of the "2-tone duct sign" and the "cork-of-wine-bottle sign."

above two typical imaging findings [1, 2], ITPN was reliably diagnosed preoperatively using EUS; thus, surgery was recommended. The resected specimen was identified with solid tumors in the dilated pancreatic

duct manifesting as soft and grayish yellow. No mucin was observed. Histopathology showed the tumor consisted of tubulopapillary structures with cylindrical cells and high grade dysplasia (► **Fig. 2a**). On immunohistochemistry, the tu-

mor was negative for MUC2, MUC6, CK-20, and CDX2 (► **Fig. 2b**), and positive for CK7, CAM5.2, and MUC-1 (► **Fig. 2c**). The Ki-67 index reached 70% (► **Fig. 2c**). The findings were consistent with an ITPN diagnosis.

As a rare pancreatic neoplasm, ITPN is very difficult to diagnose before surgery even with a combination of imaging modes [3]. We report the dynamic imaging features of multiple ITPNs of the pancreas on EUS, including the "2-tone duct sign" and "cork-of-wine-bottle sign." The limitation of this study is that EUS-guided biopsy may provide more information in the differential diagnosis between IPMN and ITPN [4, 5].

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Competing interests

The authors declare that they have no conflict of interest.

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