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Intraductal papillary neoplasm of the bile duct diagnosed by upper gastrointestinal endoscopy through a duodenobiliary fistula

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Intraductal papillary neoplasms of the bile duct (IPNBs) are a type of intraepithelial neoplasia, involving papillary growth in the bile duct. The 2019 World Health Organization (WHO) classification stated that IPNB was pathologically characterized by dilated bile ducts filled with a non-invasive papillary or villous neoplasms covering delicate fibrovascular stalks [1]. Classical IPNB was typically develops in the intrahepatic bile ducts with rich mucin production. Many cases are composed of high grade dysplasia with not infrequent microscopic foci of low/ intermediate grade dysplasia, and actually, some cases are only composed of low/intermediate dysplasia or adenoma, suggesting multi-step carcinogenesis [2]. Most cases of IPNB are diagnosed by examining surgical specimens. IPNB remains diagnostic challenge [3] [4]. Here, we report a case of IPNB, which was directly diagnosed by performing upper gastrointestinal endoscopy through a duodenobiliary fistula.

An 86-year-old female was referred to our hospital with repeated cholangitis. Contrast-enhanced CT revealed diffuse bile duct dilation. The dilation predominantly affected the left intrahepatic bile duct (IHBD) (fig.1). Fistula formation was suspected in the common bile duct and duodenum. Upper

gastrointestinal endoscopy (endoscope: GIF-H290, Olympus, Tokyo, Japan) revealed a large duodenobiliary fistula in the duodenal bulb (fig.2a). The endoscope was directly advanced from the fistula to the bile duct. The bile duct was filled with mucus (fig.2b), and multiple papillary nodules were found localized in the left IHBD (fig.2cde). An endoscopic biopsy of the nodules revealed the papillary growth of atypical cells with mucin (Fig.2f). These findings are characteristic of classical IPNBs. In the duodenal fistula, no neoplastic lesion was observed endoscopically, and only inflammatory tissue was found during a biopsy. Due to the patient's high age, follow-up observation without surgery was selected.

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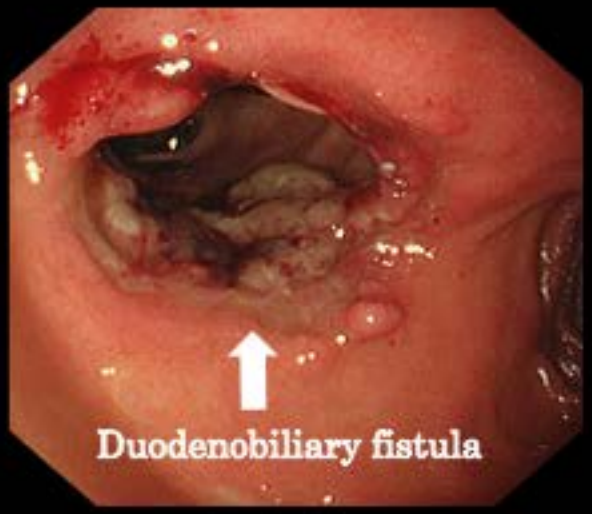
Fig1. A contrast-enhanced CT scan revealed diffuse bile duct dilation. The dilation predominantly affected the left intrahepatic bile duct.

Fig2. An upper gastrointestinal endoscopy revealed the duodenobiliary fistula in the duodenal bulb(a). The bile duct was filled with mucus (b), and multiple papillary nodules were found localized in the left IHBD (c: white light, d:

indigo carmine, e: narrow band imaging). An endoscopic biopsy of the nodules revealed the papillary growth of atypical cells with mucin(f).

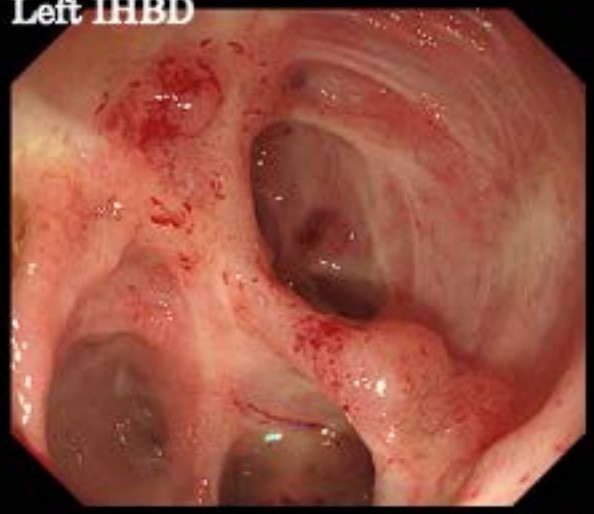

a


Duodenobiliary fistula

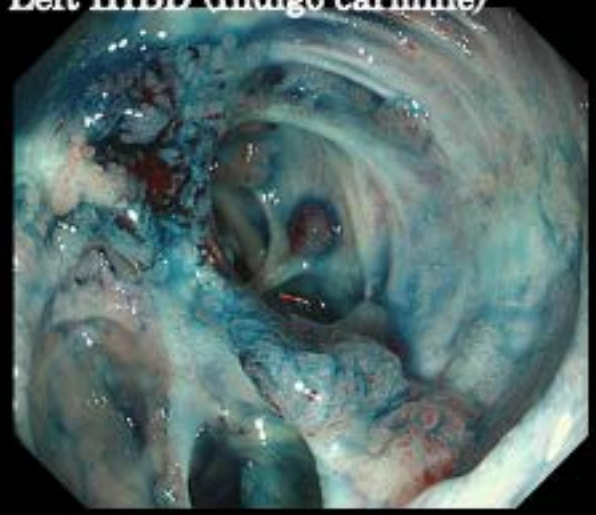
a


Left IHBD (with mucus)

b


Left IHBD

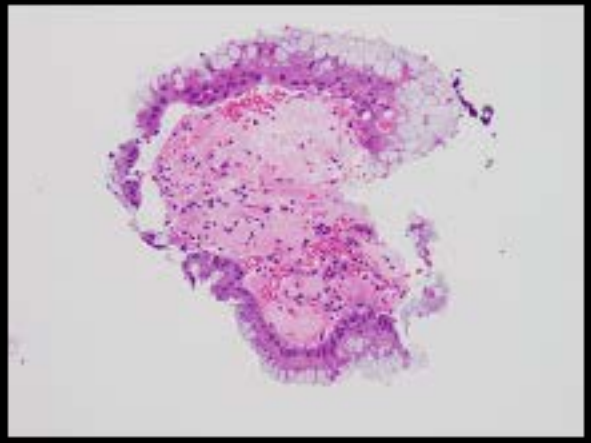
c


Left IHBD (Indigo carmine)

d


Left IHBD (NBI)

e



f