Biliary-enteric fistula is a relatively rare condition involving a communication between the biliary tree and the gastrointestinal tract [1, 2]. Parapapillary choledocho-duodenal fistula is usually asymptomatic and therefore this disorder has usually been diagnosed incidentally [3]. We report a rare case of choledocho-duodenal fistula caused by adenocarcinoma of the distal common bile duct (CBD), which was suspected on computed tomography (CT) of the abdomen and confirmed by esophago-gastro-duodenoscopy (EGD).

A 48-year-old woman presented to the emergency department with a 1-day history of epigastric pain. For the previous 2 months she had suffered intermittent epigastric pain with radiation to the back. She had no history of gallstone or gastroduodenal ulcer. She was afebrile on admission, and physical examination demonstrated epigastric tenderness. Laboratory data showed a white blood cell count of 8400/μL, an alkaline phosphatase level of 77 U/L, and normal levels of tumor markers such as carcinoembryonic antigen and cancer antigen 19–9. CT of the abdomen showed pneumobilia in both the common hepatic duct and the CBD (Fig. 1a) and a probable choledocho-duodenal fistula (Fig. 1b). EGD revealed the orifice of a choledocho-duodenal fistula 3.0 cm proximal to the papilla of Vater (Fig. 2), and a tumor in the distal CBD. Biopsy was performed and pathological examination showed moderately differentiated adenocarcinoma of the CBD. On the basis of these images, the patient was diagnosed as having advanced cholangiocarcinoma associated with a parapapillary choledocho-duodenal fistula. She underwent pylorus-preserving pancreaticoduodenectomy. Compatible with the endoscopic findings, the surgical specimen showed the orifices of the fistula and the papilla of Vater (Fig. 3a).

A papillary tumor was found in the distal CBD, where it was causing obstruction of the CBD (Fig. 3b). Pathological examination showed cholangiocarcinoma of the CBD, American Joint Committee on Cancer stage T4N0M0 (stage III). The postoperative course was uneventful.

A choledocho-duodenal fistula is an abnormal passage between the CBD and the duodenum. Common etiologies include instrumentation (iatrogenic), choledocholithiasis, and duodenal ulcer. Choledocho-duodenal fistulas associated with cancers are extremely rare, with nine cases reported in the literature; the cancers involved included ampullary cancer, duodenal cancer, gallbladder cancer, pancreatic cancer, and cholangiocarcinoma [2, 4–8]. These patients presented with epigastralgia and anorexia. CT of the abdomen often showed pneumobilia. EGD or endoscopic retrograde cholangiopancreatography (ERCP) is employed to diagnose choledocho-duodenal fistulas, and associated cancers are confirmed by biopsy.

In rare cases, the fistula is a complication secondary to an occult malignancy such as biliary adenocarcinoma. For clinical evaluation of choledocho-duodenal fistula of unknown etiology, and if occult malignancy is suspected, EGD or ERCP should be performed. The case we have reported of a patient with adenocarcinoma of the CBD causing a choledocho-duodenal fistula highlights the importance of endoscopy to diagnose choledocho-duodenal fistula and occult carcinoma of the biliary tract.

Fig. 1  Computed tomography (CT) of the abdomen showed (a) pneumobilia (black arrow) in both the common hepatic duct and the common bile duct (CBD) and (b) a possible choledocho-duodenal fistula (white arrow).

Fig. 2  Upper gastrointestinal panendoscopy revealed the orifice of a choledocho-duodenal fistula.

Fig. 3  a Compatible with the endoscopic findings, the surgical specimen showed the orifices of the fistula and the papilla of Vater. b A papillary tumor was found in the distal CBD, obstructing the duct.
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Bibliography
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