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Intraductal papillary neoplasm of the bile duct with rapidly progressive multicentric recurrence: A case report

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ABSTRACT

INTRODUCTION: Knowledge on the pattern of recurrence and prognosis of intraductal papillary neoplasms of the bile duct (IPNB) is limited. Few studies have reported IPNB recurrence in the remnant intrahepatic bile duct, which is indicative of the true multicentricity of IPNB. Herein, we report a case of IPNB with rapidly progressive recurrence in the remnant intrahepatic bile duct and review the literature for discussing the prognosis of IPNB with multicentricity.

CASE PRESENTATION: A 72-year-old male was diagnosed with IPNB in the hepatic duct of segment 3 that had spread to the left hepatic duct. The patient underwent left hepatectomy, total caudate lobectomy, and extra-hepatic bile duct resection with biliary reconstruction. Histologically, the tumor was IPNB with noninvasive adenocarcinoma with a negative surgical margin. Although dilatation of B8 and biliary enzyme elevation were observed beginning at 7–10 months postoperatively, there was no evidence of recurrence. At 17 months postoperatively, the recurrent tumor diffusely spread throughout the remnant intrahepatic bile duct. Internal drainage stents were placed within the intrahepatic bile ducts with relapsed IPNB to relieve jaundice, and a course of chemotherapy was considered. However, the patient did not receive any therapies up to his death at 21 months postoperatively because of rapid disease progression.

CONCLUSION: According to a literature review, some cases of multicentric IPNB have shown rapidly progressive recurrence and poor prognosis. We should consider multicentricity of IPNB even a few months after curative resection, and narrow examinations should also be considered.

1. Introduction

Intraductal papillary neoplasms of the bile duct (IPNB) are recognized as a subtype of biliary tumors [1] including mucin-producing bile duct tumors, papilloma, and mucinous cystic neoplasms [2]. The pattern of IPNB recurrence has been described in the literature, albeit unclearly. Particularly, reports on multicentric IPNB are rare. Herein, we report a case of IPNB with rapidly progressive recurrence in the remnant intrahepatic bile duct, indicating a possibility of multicentricity. We discuss the clinical course and prognosis and present a literature review. This work has been reported in line with the SCARE criteria [3].

2. Case presentation

A 72-year-old previously healthy man was referred to our hospital with a complaint of right upper abdominal pain. Laboratory examination revealed elevated hepatic and biliary enzyme levels: total bilirubin, 1.8 (normal range, 0.1–1.2) mg/dl; aspartate aminotransferase, 543 (normal range, 5–30) IU/l; alanine aminotransferase, 233 (normal range, 3–35) IU/l; alkaline phosphatase, 489 (normal range, 90–300) IU/l; and γ-glutamyl transpeptidase, 672 (normal range, 1–28) IU/l. Tumor marker levels (including carcinoembryonic antigen and carbohydrate antigen 19–9) were within normal limits. Contrast-enhanced computed tomography (CT) showed multilocular cystic dilatation and an enhanced mass in the hepatic duct of segment 3 (B3) (Fig. 1a). Drip infusion cholangiography CT showed a defect extending from the left hepatic duct (LHD) to the common hepatic duct (CHD) (Fig. 1b). Endoscopic retrograde cholangiography (ERC) showed an interruption in LHD above the confluence of the main hepatic ducts and a defect with mucin below (Fig. 1c). Intraductal ultrasonography revealed a 15-mm intraductal mass located between LHD and CHD

Abbreviations: CHD, common hepatic duct; CT, computed tomography; ERC, endoscopic retrograde cholangiography; IPNB, intraductal papillary neoplasms of the bile duct; LHD, left hepatic duct.

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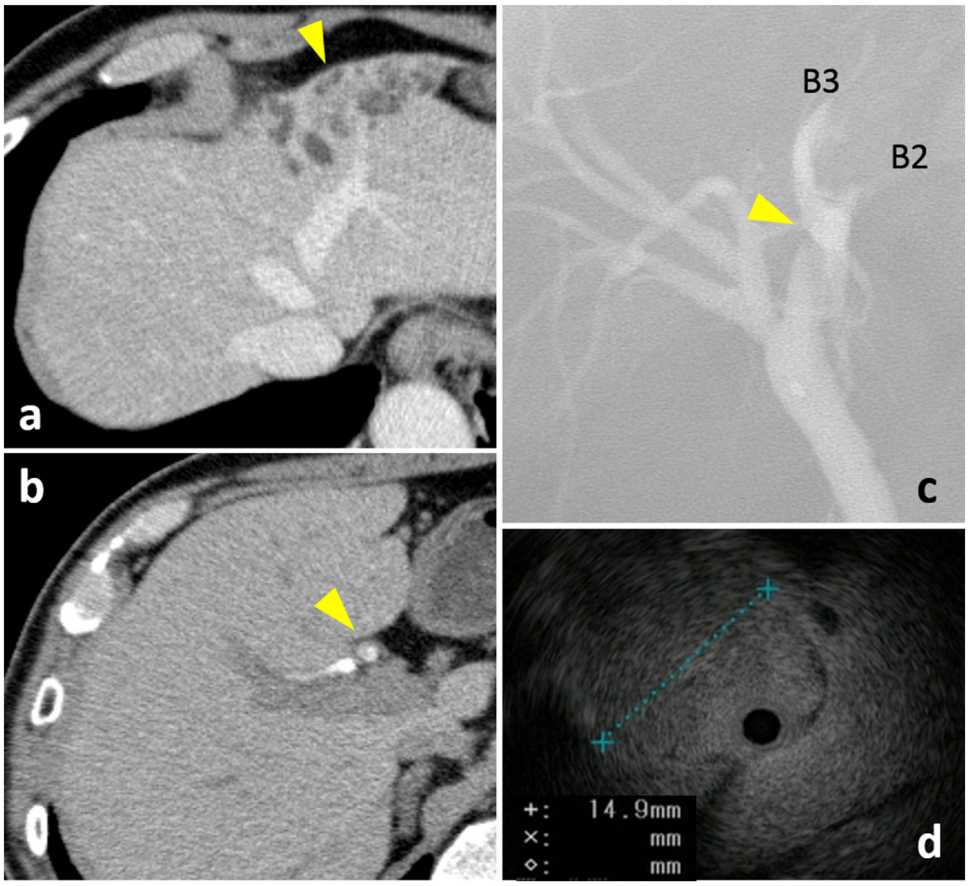

Fig. 1. Preoperative findings.
a CT showed multilocular cystic dilatation and an enhanced mass in B3 *(arrowhead)*.
b Drip infusion cholangiography CT showed a defect extending from the left hepatic duct to the common hepatic duct *(arrowhead)*.
c ERC showed an interruption in the left hepatic duct above a confluence of main hepatic ducts and a defect with mucin below *(arrowhead)*.
d Intraductal ultrasonography revealed a 15-mm intraductal growth type mass located between the left and common hepatic ducts.

(Fig. 1d). Adenocarcinoma was identified following brush cytology. According to these examinations, the patient was diagnosed with B3-dominated IPNB that spread to CHD.

The patient underwent left hepatectomy, caudate lobectomy, extra-hepatic bile duct resection with biliary reconstruction, and regional lymph node dissection. The liver parenchyma of segment 3 was remarkably atrophic. B3 was filled with tumors that appeared as irregular masses on the surface of segment 3 (Fig. 2a). The bile duct stumps were negative for cancer on frozen section examination during the surgery. Specimen cholangiography revealed that all branches of B3 had intraductal tumors. Histologically, the biliary neoplasm comprised atypical epithelial cells arranged in a highly papillary architecture with over secretion of mucin. The neoplasm spread from B3 to LHD (Fig. 2b and c). No invasive growth or lymph node metastases were observed, and the surgical margin was negative. According to these examinations, the tumor was IPNB with noninvasive adenocarcinoma. The postoperative course was uneventful; he was discharged on postoperative day 8. At 7 months postoperatively, dilatation of the hepatic duct of segment 8 (B8) was detected on CT, but it did not appear progressive. At 10 months postoperatively, his biliary enzyme levels were intermittently elevated. There was no evidence of recurrence during frequent CT or magnetic resonance imaging. At 17 months postoperatively, the patient had jaundice, and CT revealed a lesion that had diffusely spread throughout the remnant intrahepatic bile duct (Fig. 3a). Percutaneous transhepatic biliary drainage was performed, and a drainage stent was placed within the hepatic duct of segment 5 (B5), which was filled with recurrent tumors (Fig. 3b). Bile cytology revealed adenocarcinoma, confirming IPNB relapse. Drainage was insufficient because the remaining intrahepatic bile duct was filled with tumors. Internal drainage stents were placed within B5 and B8 across the anastomosis (Fig. 3c), after which the patient was discharged. Although chemotherapy was considered, owing to liver failure due to rapid progression of the disease, the patient was unable to undergo further treatments and died 21 months postoperatively.

3. Discussion

Studies have discussed unclear patterns of IPNB recurrence. Kim et al. [4] reported recurrence in 12 of 33 patients (36%) with intrahepatic IPNB. Sites of extra-hepatic recurrence were organs (40%), the peritoneum (17%), and the abdominal wall (8%). Further, the incidence of intrahepatic recurrences is unknown. Rocha et al. [5] reported recurrence in 20 of 39 patients (51%) with IPNB, including 35% with locoregional, 35% with distant, and 30% with combined recurrence. Studies have suggested underlying mechanisms of recurrence in the remnant bile duct, such as multicentricity [1,6–8], persistence of atypical epithelium [5,7], and intrabiliary dissemination [8,9]. Although cases of IPNB recurrence in the remnant bile duct have previously been thought to represent multicentric recurrence, Yokode et al. [9] described that true multicentric recurrence