Surgical Treatment of Intraductal Papillary Neoplasm of the Bile Duct: A Report of Two Cases and Review of the Literature

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Intraductal papillary neoplasm of the bile duct (IPNB) is a rare bile duct tumor characterized by intraductal papillary or villous neoplasms covered by neoplastic epithelium with fibrovascular stalks in the dilated bile ducts (1). Its true etiology remains unknown. Herein, we report two cases of IPNB that underwent surgical resection. The first case was a 66-year-old male who complained of upper abdominal pain for three years. We found obstruction of the common bile duct and dilation of the intrahepatic and extrahepatic bile ducts after MRCP. Laparoscopic hepatic segmentectomy (S2, S3, S4), resection of the common bile duct, cholecystectomy, and hepaticojejunostomy were performed. The second case was a 67-year-old male with asymptomatic dilation of the intrahepatic duct. The patient underwent robot-assisted laparoscopic hepatic segmentectomy (S5, S6, S7, S8), resection of the common bile duct, cholecystectomy, and hepaticojejunostomy were performed. The second case was a 67-year-old male with asymptomatic dilation of the intrahepatic duct. The patient underwent robot-assisted laparoscopic hepatic segmentectomy (S5, S6, S7, S8), resection of the common bile duct, cholecystectomy, and hepaticojejunostomy and cholecystectomy.

Keywords: surgical treatment, intraductal papillary neoplasm of the bile duct, mutation, clinical features, prognosis

BACKGROUND

According to the 2019 World Health Organization (WHO) classification of digestive system tumors, intraductal papillary neoplasm of the bile duct (IPNB) is a rare tumor in the bile duct that is characterized by intraductal papillary or villous neoplasms covered by neoplastic epithelium with fine fibrovascular stalks in the dilated bile ducts (1, 2). IPNB is mainly reported in Far Eastern countries where hepatolithiasis and clonorchiasis are endemic (1). Based on a multicenter analysis, malignant features of tumors are observed in 43% of IPNB cases, and patient prognoses for malignant lesions are worse than for noninvasive lesions (3). Therefore, early identification and resection of lesions are significant, even in asymptomatic patients.

Here, we report two cases of surgical treatment of IPNB with a review of relevant literature; perhaps they could provide some information to understand this rare disease.
CASE PRESENTATION

Case 1
A 66-year-old man was referred to our hospital due to abdominal pain. Three years ago, he had presented with right upper quadrant abdominal pain, which worsened after meals, and the pain was more frequent in recent months. He had no noteworthy medical or family history. Physical examination was unremarkable.

Enhanced computed tomography (CT) at our hospital revealed interruption of the intrapancreatic common bile duct (Figure 1B) and dilatation of the extrahepatic and intrahepatic bile duct, which was especially obvious in the left lobe of the liver (Figure 1A). To evaluate dilatation of the bile duct, magnetic resonance cholangiopancreatography (MRCP) was performed. MRCP showed dilatation of the extrahepatic and intrahepatic bile ducts, irregular dilatation and thickening of the bile duct in the left lobe of the liver (Figures 1C–H). At the same time, there were localized nodular prominences in the bile duct lumen. Laboratory values on admission were as follows: tumor markers, alpha-fetoprotein (AFP) 2.6 ng/mL (normal range 0.00-8.78 ng/mL), ferritin (Fer) 153.96 ng/mL (normal range 21.80–274.66 ng/mL), carcinoembryonic antigen (CEA) 2.57 ng/mL (normal range 0.00–5.00 ng/mL), and cancer antigen 19-9 (CA199) 47.85 U/mL (normal range 0.00–37.0 U/mL). A routine blood examination was not abnormal. The level of γ-glutamyltranspeptidase (γ-GTP) was elevated, 109 U/L (normal range 7–49 U/L) (Table 1).

After the relevant preoperative examination was completed, laparoscopic hepatic segmentectomy (S2, S3, S4), resection of the common bile duct, cholecystectomy, and hepaticojejunostomy were performed. On intraoperative cholangioscopy, we found the left hepatic duct filled with mucus and papillary protrusions adhering to the surface of the left hepatic duct (Figure 1I). No abnormalities were detected in the common bile duct. Laparoscopic hepatic segmentectomy (S2, S3, S4), resection of the common bile duct, cholecystectomy, and hepaticojejunostomy were performed. The bile duct was filled with greenish-yellow mucus-like sludge (Figures 1J, K). Microscopically, the intrahepatic bile duct, common bile duct and cystic duct were lined by papillary growth neoplasia with high-grade intraepithelial neoplasia (Figures 1L, M). Gene detection was performed using DNA extracted from paraffin-embedded bile ducts. Gene detection revealed KRAS codon 12 (p.G12F) mutation, and the mutation frequency was 62.9% (Figure 1N).

The postoperative course was uneventful, and the patient was discharged from the hospital on the 7th postoperative day. The patient had no recurrence and no complications for 8 months after surgery.

Case 2
A 67-year-old male with diabetes mellitus was admitted to our hospital due to finding dilatation of extrahepatic and intrahepatic bile ducts by accident during a routine medical examination 15...
TABLE 1 | Laboratory data before and after the surgery.

| Tumor markers | Case 1 | Case 2 | Normal range |
|---------------|--------|--------|--------------|
| Pre-operation | Post-operation (6 days) | Post-operation (8 months) | Pre-operation | Post-operation (10 days) | Post-operation (6 months) |
| **AFP** | 2.6 | – | – | 1.97 | – | 1.54 | 0.00-8.78 ng/mL |
| **Fer** | 153.96 | – | – | 288.81 | – | 195.62 | 21.80-274.66 ng/mL |
| **CEA** | 2.57 | – | – | 4.91 | – | 2.65 | 0.00-5.00 ng/mL |
| **CA199** | 47.85 | – | 28.65 | 95.00 | – | 35.55 | 0.00-37.00 U/mL |
| **Routine blood tests** | | | | | | | |
| **WBC** | 4.21 | 7.15 | 5.74 | 9.81 | 9.02 | – | 3.50-9.50×10⁹/L |
| **RBC** | 4.69 | 3.84 | 4.46 | 5.74 | 4.22 | – | 4.30-5.80×10¹²/L |
| **Hb** | 139 | 118 | 131 | 163 | 122 | – | 130-175g/L |
| **PLT** | 216 | 170 | 149 | 171 | 139 | – | 125-350×10⁹/L |
| **NEU%** | 65.3 | 69.7 | 62.4 | 77.0 | 70.4 | – | 40.0-75.0% |
| **Liver function tests** | | | | | | | |
| **ALB** | 45 | 37 | 44 | 41 | 34 | – | 35-55 g/L |
| **ALT** | 17 | 57 | 17 | 20 | 78 | – | 5-40 U/L |
| **AST** | 19 | 24 | 21 | 17 | 27 | – | 5-40 U/L |
| **ALP** | 90 | 58 | 42 | 104 | 68 | – | 40-150 U/L |
| **γ-GTP** | 143 | 92 | 35 | 196 | 84 | – | 7-49 U/L |

Intraoperative cholangioscopy was performed to evaluate the common bile duct. The common bile duct wall was rough and full of white floculus (Figures 2G, H). Therefore, the patient underwent robot-assisted laparoscopic hepatic segmentectomy (S5, S6, S7, S8), resection of the common bile duct, hepaticojejunostomy and cholecystectomy. Grossly, the intrahepatic bile duct lumen was filled with yellow, gelatinous, sticky mucous masses. The diameter of the largest one was 18 mm. (Figures 2I, J) Pathologic findings were as follows: Intraductal papillary neoplasm of the bile duct with low-grade intraepithelial neoplasia, focal high-grade intraepithelial neoplasia; a large number of lymphocytes aggregated in the portal area with extensive bile duct proliferation (Figures 2I, J). The patient refused gene detection for financial reasons. The patient’s postoperative course was uneventful. The patient was discharged from our department 13 days after surgery. The patient was followed up for 6 months after the operation, and there were no signs of recurrence. (Figure 2K).

**DISCUSSION**

Intraductal papillary neoplasm of the bile duct (IPNB) is a rare bile duct tumor characterized by intraductal papillary or villous neoplasms covered by neoplastic epithelium with fine fibrovascular stalks in the dilated bile ducts (1).

IPNB is mainly found in patients in Eastern countries, such as Japan, and Korea, where hepatolithiasis and clonorchiasis, which are known to be major risk factors for IPNB, are endemic (4). IPNB shows a slight male predominance, and most patients are between 50 and 70 years of age is reported (4, 5). However, the pathogenesis and nature of IPNB are still unclear. It is likely caused by cholestasis, biliary tract infection, and biliary tract cancer. Studies have found a mechanism for biliary tract cancer due to the progression of chronic inflammation to multistage carcinogenesis and eventually hyperplasia-dysplasia-carcinoma (6). Furthermore, chronic inflammation induces the production of reactive oxygen species and reactive nitrogen species, resulting in DNA damage, which plays an important role in carcinogenesis (7). IPNB symptoms include recurrent and intermittent abdominal pain, cholangitis, and jaundice. However, some patients are asymptomatic (3, 8). Histological types of IPNB have been classified into the following four types: gastric, intestinal, pancreaticobiliary, oncocytic, and IPMN, depending on morphologic appearance and mucin staining properties (9). GNAs and KRAS mutations detected in 50% and 46.2% of IPNBs are common in IPNBs. KRAS plays an important role in regulating cell growth and differentiation (10). A recent study has shown that KRAS mutation was detected in one
lymph nodes (4). Recently, new approaches, such as RFA and APC, have been helpful for patients who are not candidates for surgery due to age or physical condition (17, 18). Liver transplantation and hepatectomy and segmental pancreatectomy. A total of 7.5% of patients underwent hepatectomy, extrahaepatic bile duct resection, and hepaticojejunostomy. Five percent of patients underwent hepatectomy and segmental pancreatectomy. A total of 7.5% of patients underwent hepatocellular carcinoma. Immunohistochemical data were available for 26 cases, from which we found that CK7 (42.3%), MUC5AC (53.8%) and MUC6 (53.8%) expression was common in the IPNB. Based on the recurrence-free survival (RFS) reported in 23 cases, we found that the range of RFS was 4.0-39.0 months, with a median of 14.0 months (Table 3).
On further review of the literature, we could conclude that IPNB mainly behaves as in imaging solid mass, cystic lesion and dilation of the bile duct in imaging. Surgical resection is the major treatment for patients in fine condition, and interventional therapies such as RFA and APC are good choices for palliative treatments. Although most IPNB cases are high-grade intraepithelial neoplasia or invasive carcinoma in microscopically, surgical resection could make patients get satisfactory prognosis.

CONCLUSION

In summary, we report two cases of IPNB, a rare tumor of the hepatobiliary system, and analyze published case reports about IPNB. Since IPNB has a high potential for transforming into an invasive lesion, R0 surgical resection is preferred. At the same time, RFA and APC may be new palliative approaches for treating IPNB.

DATA AVAILABILITY STATEMENT

The datasets presented in this study can be found in online repositories. The names of the repository/repositories and accession number(s) can be found in the article/supplementary material.

AUTHOR CONTRIBUTIONS

BL designed the case report. ZL, QL, and WT participated in the operation and management of the patients. BL, ZM, and ML prepared radiological and histology figures. BL, ZL reviewed the literature and drafted the article. All authors contributed to the article and approved the submitted version.

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