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Case Report
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Choledochal Cholangiocarpal Adenoma with Intrahepatic Bile Duct Stones: A Case Report

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Abstract

This case report describes a 58-year-old female patient with choledochal cholangiocarcinoma and intrahepatic bile duct stones. The patient had a history of hepatic left lobectomy and ERCP for bile duct stones. Imaging revealed bile duct dilation, choledochal stones, and a neoplastic lesion. She underwent left hemihepatectomy, choledochotomy, and Roux-en-Y choledochojejunostomy. Histopathology confirmed choriocapillary adenoma with high-grade intraepithelial neoplasia. The patient recovered well with no recurrence on follow-up. The case highlights the importance of early diagnosis and surgical resection for this rare tumor.

Keywords: Hepatoportal tumor; Extrahepatic biliary adenoma; Roux-en-Y choledocho-jejunostomy

Case Information

Patient: Female, 58 years old. She was admitted to Sinopharm Dongfeng General Hospital ("our hospital") with the chief complaint of epigastric discomfort for 1 week, more than 4 years after hepatic left lobectomy and more than 2 years after ERCP. Past medical history: The patient was hospitalized in our hospital 4+ years ago for intrahepatic and extrahepatic bile duct stones, and underwent laparoscopic hepatic left lobectomy + laparoscopic choledochotomy with cholangioscopy + T-tube drainage + laparoscopic cholecystectomy. Two years ago, the patient was diagnosed with choledochal stones, and treated with endoscopic retrograde cholangiopancreatography, duodenal papillary muscle incision lithotripsy + endoscopic nasocholestracheal drainage. The possibility of papillary tumor in the bile duct was high. Post-admission examination: The abdomen was flat and soft; the abdominal muscle was soft, with no mass, obvious tenderness, or rebound tenderness. The liver and spleen were not palpable under the rib cage. There was no percussion pain in the liver region or right renal region; there were no shifting dullness, and bowel sounds were normal. Liver function tests after admission: total bilirubin 23.9 μmol/L, direct bilirubin 12.4 μmol/L, alanine aminotransferase 90 U/L, aspartate aminotransferase 47 U/L. Magnetic Resonance Cholangio-Pancreatography (MRCP) showed: 1) postoperative review of intrahepatic and extrahepatic choledochotomy: marked dilatation of intrahepatic bile ducts and common bile ducts, choledochal stone in the right hepatic lobe with mild cholangitis; 2) residual flocculent shadow in the left hepatic duct (Figure 1a). Abdominal enhanced CT showed: flocculent soft tissue shadow in the dilated bile ducts of the hepatoportal region (Figure 1b), suggesting a neoplastic lesion (possibly intrahepatic papillary mucinous neoplasm?). Circumscribed slightly hyperdense shadow was noted within the dilated bile duct in the right hepatic lobe; clinical correlation was recommended for further characterization. Diffuse dilatation of intrahepatic and extrahepatic bile ducts was observed, with a small amount of pneumobilia in the hilar region. Preoperative diagnosis: bile duct occupancy; choledocholithiasis complicated with cholangitis; postoperative status of hepatic left outer lobe resection; postoperative status of cholecystectomy; postoperative status of choledochotomy; postoperative status of ERCP; hypertension. Based on the patient's condition, infection control and liver function improvement were implemented: combined anti-infective therapy and hepatoprotective drugs (magnesium isoglycyrrhizinate) were administered. After treatment, the patient's body temperature normalized, abdominal pain symptoms were alleviated, and liver function indices showed improvement. Nutritional support and comorbidity management were simultaneously provided: Due to chronic disease-related wasting, preoperative nutritional risk screening was performed, and enteral nutritional support was given to improve nutritional status. Additionally, a cardiology consultation was requested to adjust antihypertensive regimens for maintaining the stability of perioperative blood pressure. Comprehensive assessment of vital organ functions was completed: via electrocardiogram, cardiac ultrasound, and pulmonary function tests,

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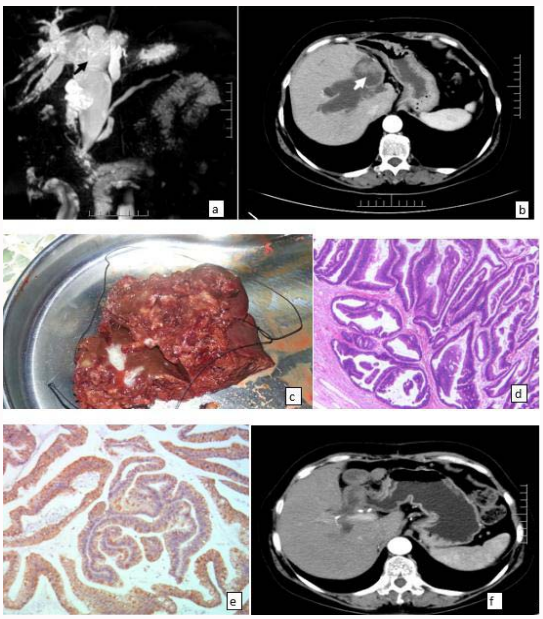

Figure 1: Shows the patient's preoperative imaging findings, postoperative specimen, histopathology results, and postoperative imaging findings.
a: Preoperative MRCP showed dilated bile ducts and residual flocculent shadows in the left hepatic duct;
b: Enhanced CT showed flocculent soft tissue shadows in the dilated bile ducts in the hepatic hilar region;
c: Gross left hemiliver pathology specimen;
d: Postoperative histopathology revealed the tissue to be a choroidal tubulovillous adenoma (grade II~III) with high-grade intraepithelial neoplasia. Hepatocellular edema was noted, and focally lymphocytic infiltration was observed in the interstitium (HE staining × 20);
e: Positive staining for broad-spectrum cytokeratin (immunohistochemical staining × 40);
f: Enhanced CT of the epigastric region at 4 months postoperatively showed postoperative changes following resection of the left hepatic lobe, resection of the choledochotomy, choledochoenteric anastomosis, and enteroenteric anastomosis.

the patient's cardiopulmonary tolerance for surgery was confirmed. Preoperative discussion and plan formulation: A multidisciplinary team (MDT) discussion involving hepatobiliary surgery, anesthesiology, radiology, and pathology departments was organized. Based on imaging findings and medical history, the team concluded a high probability of neoplastic lesions located in the left hepatic duct involving the hepatic hilum, indicating surgical indications. Later, the patient underwent left hemihepatectomy + choledochotomy + Roux-en-Y choledocho-jejunostomy. Intraoperatively, dilatation of the left intrahepatic bile duct and common bile duct was noted, and a tumor with a diameter of about 5 cm was palpated inside, with soft texture, slightly reduced mobility, and unclear borders. Total intraoperative hemorrhage was approximately 200 ml. Postoperative gross examination of the specimen showed that the left hepatic duct measured 3.0 cm in length and 3.5 cm in diameter, with a bulging mass within the duct measuring 3.0 cm × 3.0 cm × 2.5 cm, appearing as a grayish-reddish solid mass, with soft texture and unclear boundaries. The liver section showed a dilated bile duct measuring 0.5~1.3 cm in diameter, with stones in the lumen. Also identified was common bile duct tissue measuring approximately 4.0 cm in length and 1.5 cm in diameter; no lymph nodes were identified in the adipose tissue surrounding the liver (Figure 1c).

Postoperative histopathological findings: (left hepatic duct and adjacent intrahepatic bile duct) choriocapillary adenoma (grade II~III) with high-grade intraepithelial neoplasia (Figure 1d), tumor size 3.0 cm × 3.0 cm × 2.5 cm. Choriocapillary adenoma (grade III) with high-grade intraepithelial neoplasia at the surgical margin of the left hepatic duct. Pigmented gallstones were seen attached to the local duct wall; no neoplastic lesion was observed; the intrahepatic bile ducts (left hemihepatic region) were dilated, with stones seen in the lumen.

Immunohistochemical staining results: CKpan(+) (CKpan is a broad-spectrum cytokeratin that labels almost all epithelia, including overlying epithelia and glandular epithelia, and is expressed in various squamous carcinomas, adenocarcinomas, transitional cell carcinomas, and small cell carcinomas), p53 (variable intensity), Ki-67 (60% positive), smooth muscle actin (-), Desmin (-), CD34 (no tumor thrombus in the blood vessels of the examined tissues), D2-40 (no tumor thrombus in the lymphatic vessels of the examined tissues), HBsAg (-), and HBcAg (-). The patient had an uneventful postoperative recovery and was discharged on the 14th postoperative day. When the patient returned to the hospital at 4 months postoperatively, the enhanced CT of the upper abdomen showed: postoperative changes in the left hepatic lobe, following choledochotomy, postoperative changes at the bilioenteric anastomosis and enteroenteric anastomosis (Figure 1e), prominent peripheral lymph nodes, postoperative changes in the right abdominal wall, widening of the bile ducts in the right hepatic lobe, and a small amount of pneumatization in the intrahepatic bile ducts. The patient was in good general condition and denied abdominal pain or distension.

Discussion

Choledochal cholangiocarpal adenoma is extremely rare in clinical practice and has malignant tendency, which can progress to adenocarcinoma [1]. Its etiology remains unclear, but it may be related to the prolonged stimulation of gallstones, inflammation, and other factors that induce bile duct epithelial hyperplasia and tissue ectasia, further leading papillary hyperplasia and papilloma; it may be also related to mutations of BRAF ^{V600E} and BRAF^{V600E23} genes [2]. Choledochal cholangiocarpal adenoma exhibits significant cellular heterogeneity, and the complex interactions between tumor cells and neural cells in the tumor microenvironment serve as key mechanisms driving perineural invasion (PNI) [3]. This heterogeneity may also exist during the carcinogenesis of villous adenoma, as intraductal papillary neoplasm of the bile duct (IPNB)–a precancerous lesion–demonstrates pathological features: cancer cells originate within the bile duct lumen, and subsequently invade the surrounding liver tissue [4]. Its pathogenesis is associated with abnormal bile duct branching and tubule formation regulated by fibroblast growth factor 10 (FGF10). Induced FGF10 expression in animal models can recapitulate the progression of IPNB [5]. The patient in this case had previously undergone bile duct surgery for bile duct stones. The current condition did not exclude the formation of papillary (cholangiocarpal) adenomas due to repeated irritation from bile duct stones and cholangitis. Cholangiocarpal adenomas are usually asymptomatic; however, if located in the hilar region, they can lead to obstructive jaundice. The common symptoms of cholangiocarpal adenomas include intermittent pain, dyspepsia, weight loss, nausea, vomiting, malaise, and fever. In an imaging-based study [3], 60% of patients with cholangiocarpal adenomas were found to have dilated bile ducts. According to Muro et al. [4], brush cytology is a common technique for the initial examination. Since the diagnosis of this disease is complex, it is important to comprehensively analyze and evaluate the patient's clinical symptoms, serological findings, and

imaging parameters to determine the optimal treatment strategy. Although histologically benign, cholangiocarpal adenomas are now recognized to have a tendency for malignant transformation and to grow along the mucosal surface of the bile ducts, potentially developing into adenocarcinomas. As long as the lesion is resectable and the patient can tolerate the procedure, complete resection remains the preferred treatment option. In a report by Zhou Haixiang [5], ERCP provides good relief of clinical symptoms in patients on a nonsurgical treatment regimen, but carries a risk of recurrence. Surgery is indicated if malignancy is suspected or tumor >2 cm. In our patient, the tumor was located in the middle and upper part of the common bile duct, and was complicated by left residual intrahepatic stones. Therefore, left hemihepatectomy + choledochotomy + Roux-en-Y hepaticojejunal anastomosis was performed, achieving a favorable outcome. Pancreaticoduodenectomy is indicated in cases involving the distal common bile duct where malignancy cannot be excluded. In addition, some investigators [6] believe that local resection+ hepaticoduodenal ligament lymph node dissection is also indicated for lesions with malignant tendency in the middle and lower segments of the common bile duct, and that liver transplantation may be a treatment option for patients with advanced disease. Current short-term follow-up studies [7-10] show that cholangiocarpal adenomas rarely recur after surgery. In conclusion, cholangiocarpal adenoma is a rare benign tumor of the extrahepatic bile ducts, and early diagnosis and resection are of great importance. For this patient, whose tumor was located in the middle and upper part of the common bile duct, left hemihepatectomy + choledochotomy + Roux-en-Y hepaticojejunal anastomosis was performed with reasonable surgical approach. The operation proceeded smoothly without serious perioperative complications, and the patient achieved a favorable prognosis.

Statement

Conflict of interest statement: All authors of this article have read and understood the policy statement of the Chinese Journal of Basic and Clinical Surgery, and we have no competing interests.

Author Contribution Statement: Hao Li was responsible for case collection, data analysis, and writing the paper; Wenbo Zhou was responsible for formulating the writing ideas, revising the paper, and finalizing it.

Ethical Statement: This study was approved by the Ethics Committee of tsinopharm Dongfeng general Hospital (Approval No. LW-2024-022).

Conflict of Interest

The authors declares no conflict of interest related to this work.

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