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Synchronous double primary cancers of the extrahepatic bile duct: A case report and literature review

Takeshi Nishi[a,b,*], Yoshitoshi Sato[b], Takuya Hanaoka[c], Takuya Takahashi[d], Hiroshi Miura[d], Kenji Takubo[b]

[a] Department of Digestive and General Surgery, Shimane University Faculty of Medicine, Shimane, Japan
[b] Department of Surgery, Matsue Red Cross Hospital, Shimane, Japan
[c] Department of Gastroenterology, Matsue Red Cross Hospital, Shimane, Japan
[d] Department of Pathology, Matsue Red Cross Hospital, Shimane, Japan

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ABSTRACT

BACKGROUND: Double cancers of the biliary tract system are rare. Most of these cancers are synchronous double cancers of the gall bladder and bile duct, associated with pancreaticobiliary maljunction (PBM). Synchronous double cancers of the extrahepatic bile duct without PBM are especially rare, and only 4 cases have been reported.

CASE PRESENTATION: A 78-year-old woman was admitted to our hospital for examination of hyperbilirubinemia and liver dysfunction. Contrast-enhanced abdominal computed tomography, Magnetic resonance cholangiopancreatography and endoscopic retrograde cholangiopancreatography revealed 2 stenotic regions in the common bile duct: at its junction with the cystic duct and in the distal bile duct. No findings suggested PBM, such as a markedly long common channel. The diagnosis based on endoscopic brush cytology from both stricture portions was adenocarcinoma. The patient had a pylorus-preserving pancreaticoduodenectomy with regional lymph node resection. Macroscopically, there were 2 stenotic regions at the cystic duct junction and in the distal bile duct. Microscopically, the tumor at the junction of the cystic duct was a well-to-moderately differentiated adenocarcinoma. On the other hand, the tumor of the distal bile duct was a poorly differentiated adenocarcinoma. There was no evidence of communication between these 2 cancers.

CONCLUSION: Double cancers of the extrahepatic bile duct without PBM are very rare. Therefore, an accurate diagnosis prior to surgery is necessary. Furthermore, this rare condition seems to be associated with a poor prognosis.

1. Background

Double cancers of the biliary tract system are rare. Most reports are of synchronous gall bladder and bile duct cancers, and cases are often associated with pancreaticobiliary maljunction (PBM) [1]. Synchronous double cancers of the extrahepatic bile duct without PBM are especially rare, and to date only 4 cases have been reported in the English literature [2–5]. We herein report a case of synchronous double primary cancers of the extrahepatic bile duct and review the relevant literature. This study has been reported in line with the SCARE criteria [6].

2. Case presentation

A 78-year-old woman visited her local doctor with appetite loss, abdominal fullness, and dark urine. She had suffered from a drug-induced hepatitis due to an agricultural chemical in youth and she was receiving treatment for hypertension, hyperlipidemia, and vertigo. On admission, the patient had jaundice, and there was no palpable mass in the abdomen. Laboratory test results revealed hyperbilirubinemia (total bilirubin 7.6 mg/dl; normal range 0.3–1.3 mg/dl, direct bilirubin 6.1 mg/dl; normal range 0.0–0.4 mg/dl) and high serum levels of several liver enzymes (aspartate aminotransferase 240 U/L; normal range 11–34 U/L, alanine aminotransferase 347 U/L; normal range 7–34 U/L, alkaline phosphatase 1426 U/L; normal range 110–340 U/L, r-glutamyltranspeptidase 1097 U/L; normal range 5–55 U/L). Carbohydrate antigen 19-9 levels were high (328 U/ml;

Abbreviations: PBM, pancreaticobiliary maljunction; CT, computed tomography; MRCP, magnetic resonance cholangiopancreatography; ERCP, endoscopic retrograde cholangiopancreatography.

* Corresponding author at: 89-1 Enyacho, Izumo, 693-8501, Japan.
E-mail addresses: nishiken@med.shimane-u.ac.jp (T. Nishi), ys8312002@yahoo.co.jp (Y. Sato), manekineko0624@yahoo.co.jp (T. Hanaoka), matsue.byouri@gmail.com (T. Takahashi), hmiu3@yahoo.co.jp (H. Miura), k.tkb50@yahoo.co.jp (K. Takubo).

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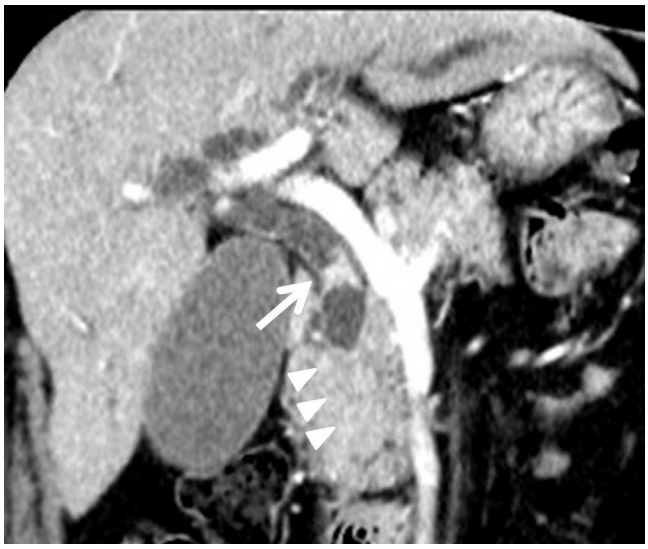

Fig. 1. Coronal image of contrast-enhanced computed tomography of the abdomen. The common bile duct wall in the junction of cystic duct is thickened (arrows). The common bile duct does not trace until the papilla of Vater, which suggests that there is stenosis in the distal bile duct (arrowheads).

normal range 0–37 U/ml), and carcinoembryonic antigen levels were normal. An abdominal ultrasonography showed a dilation of the intrahepatic bile duct in both liver lobes and the common bile duct measuring 11 mm in diameter. No tumors were detected in the common bile duct or in the head of the pancreas. A contrast-enhanced abdominal computed tomography (CT) scan revealed thickening of the common bile duct wall at its junction with the cystic duct and dilation of the peripheral bile duct from this point. On CT, another obstruction was found in the distal portion of bile duct. There was no lymph node swelling around the extrahepatic bile duct (Fig. 1). Magnetic resonance cholangiopancreatography (MRCP) revealed 2 stenotic regions in the common bile duct. One portion was at its junction with the cystic duct and the other was in the distal bile duct (Fig. 2a). Endoscopic retrograde cholangiopancreatography (ERCP) showed similar findings to the MRCP (Fig. 2b). There were no findings indicative of PBM, such as a long common channel, in either the MRCP or ERCP examinations. The diagnosis based on endoscopic brush cytology from the upper stricture of the common bile duct was adenocarcinoma and the diagnosis from the distal stricture was suspicious of adenocarcinoma. The patient had a pylorus-preserving pancreaticoduodenectomy with regional lymph node resection. The postoperative course was uneventful, with the exception of "biochemical leak" postoperative pancreatic fistula [7] and chylous ascites (Grade I according to the Clavien-Dindo classification [8]). The patient was discharged on postoperative day 26.

Macroscopically, there were 2 stenotic regions with rough mucous membrane: in the common bile duct and cystic duct junction and in the distal bile duct (Fig. 3). Microscopically, the tumor at the junction of the cystic duct was a well-to-moderately differentiated adenocarcinoma. The tumor cells had spread to the middle bile duct and invaded the under layer of the serosa (Fig. 4a). On the other hand, the tumor of the distal bile duct was a poorly differentiated adenocarcinoma (Fig. 4b). The tumor cells had invaded the pancreas and the fatty tissue outside the bile duct. Sections of the tumor had neuroendocrine carcinoma-like features, but immunohistochemical staining for neuroendocrine tumor markers, such as CD56, chromogranine A, and synaptophysin, was negative. There was no communication between the 2 cancers (Fig. 4c) and there was no lymph node metastasis. According to the TNM classification [9], the tumor of the cystic duct junction was classified as pT1N0M0,

Fig. 2. (a) Magnetic resonance cholangiopancreatography; There are 2 stenotic portions in the common bile duct. One portion is in the junction of the cystic duct (arrows), and the other is in the distal bile duct.The common channel is of normal length. (b) Endoscopic retrograde cholangiopancreatography; There are 2 stenotic portions in the common bile duct. One portion is in the middle bile duct (arrows), and the other is in the distal bile duct (arrowheads).The common channel is of normal length.

Stage IA, and the tumor of the distal bile duct was classified as pT3N0M0, Stage IIA.

The patient did not want to receive adjuvant chemotherapy, and 8 months later, a CT scan revealed multiple liver metastases. The patient then received chemotherapy with gemcitabine and cisplatin, and died 18months after the operation.

3. Discussion

Double cancers of the biliary system sometimes occur in PBM patients [1]. The mechanism of carcinogenesis in PBM appears to be related to the persistent reflux of pancreatic juice into the biliary