Case report

Extrahepatic biliary cystadenocarcinoma arising from
the left hepatic duct

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Abstract: A 54-year-old man, who had no clinical symptoms, underwent a routine health checkup at our hospital. Abdominal ultrasonography disclosed a well demarcated tumor containing a solid portion occupying the dilated left hepatic duct and a cystic portion expanding into the parenchyma of the left hepatic lobe, with mild dilatation of the intrahepatic bile ducts. These findings were later confirmed by computed tomography (CT) and magnetic resonance imaging. Endoscopic retrograde cholangiography revealed a complete defect at the level of the left hepatic duct, while drip infusion cholangiographic-CT (DIC-CT) disclosed a defect of the left hepatic duct only, with the distal portions of the left intrahepatic ducts being visualized on the image. Hepatic angiography revealed light stains in the solid portion in the parenchymal phase. At left lobectomy, a multiloculated polyp-like tumor was found arising from the left hepatic duct and expanding into the parenchyma of the left hepatic lobe. Microscopically, all the lining cells in the cysts and the tumor cells in the solid portion showed the features of papillary adenocarcinoma. In this patient with extrahepatic biliary cystadenocarcinoma, DIC-CT was useful in identifying the site of origin of the tumor, and hepatic angiography was also useful in differentiating this rare malignant tumor from benign cystadenoma.

Key words: extrahepatic biliary cystadenocarcinoma, DIC-CT, hepatic angiography

Introduction

Reports of intrahepatic biliary cystadenocarcinomas have been increasing, because of advances in imaging diagnoses and due to the establishment of the character of this tumor.1–3 Reports of extrahepatic biliary cystadenocarcinomas are comparatively fewer.4–8 We report here a case of biliary cystadenocarcinoma arising from the left hepatic duct in a patient without jaundice, which is the most common symptom of extrahepatic biliary cystadenocarcinoma.

Case report

A 54-year-old man with no clinical symptoms underwent a routine health checkup at which a solid and cystic tumor 4.5 cm in diameter, situated in the left hepatic duct, was detected incidentally by abdominal ultrasonography (US) (Fig. 1). His liver function test results were entirely normal, as were serum levels of carcinoembryonic antigen, carbohydrate 19-9 antigen, and alpha-feto protein. Abdominal computed tomography (CT) and magnetic resonance imaging demonstrated a well demarcated tumor containing a solid portion occupying the dilated left hepatic duct and a cystic portion expanding into the parenchyma of the left hepatic lobe, with mild dilatation of the intrahepatic bile ducts. Endoscopic retrograde cholangiography (ERC) revealed a complete defect at the level of the left hepatic duct (Fig. 3). A three-dimensional drip infusion cholangiographic-CT (DIC-CT) image showed a defect at the left hepatic duct only, and the distal portions of the left intrahepatic ducts were visualized on the image (Fig. 4). Hepatic angiography revealed light stains in the solid portion in the parenchymal phase (Fig. 5). He underwent a left lobectomy. A multiloculated polyp-like tumor was found arising from the left hepatic duct and expanding into the parenchyma of the left hepatic lobe (Fig. 6). Mucinous fluid filled each loculus. Part of the left hepatic duct was tumor free, and its biliary flow was preserved. Microscopically, all the
Fig. 1. Ultrasonography reveals a solid and cystic mass in the dilated left hepatic duct

Fig. 2a,b. Abdominal computed tomographic scan demonstrates a well demarcated tumor containing a solid portion occupying the dilated left hepatic duct and b a cystic portion expanding into the parenchyma of the left hepatic lobe, with mild dilatation of the intrahepatic bile ducts

The lining cells in the cysts showed strong cellular and nuclear atypia (Fig. 7). The solid portion presented features consistent with dense proliferated papillary adenocarcinoma.

Fig. 3. Endoscopic retrograde cholangiography reveals a complete defect at the level of the left hepatic duct

Fig. 4. Three-dimensional drip infusion cholangiographic computed tomographic image discloses a defect of the left hepatic duct only, and the distal portions of the left intrahepatic ducts are visualized on the image

The patient has been free of tumor recurrence for 2 years since the surgery.

Discussion

Extrahepatic biliary cystadenocarcinoma is a rare malignant tumor. To the best of our knowledge, only four