Endobiliary hepatocellular carcinoma: A rare tumor mimicking cholangiocarcinoma

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LETTER / *Abdominal imaging*

Endobiliary hepatocellular carcinoma: A rare tumor mimicking cholangiocarcinoma

Keywords Hepatocellular carcinoma; Endobiliary growth; MR imaging

Dear Editor,

Invasion of main bile ducts by hepatocellular carcinoma (HCC) has an incidence ranging between 0.53% and 9% [1,2]. Patients typically present with jaundice and right upper quadrant pain, which may be related either to migration of tumor fragments or hemobilia by intratumoral bleeding. In general, the associated HCC is clearly visible and biliary involvement corresponds to direct invasion by the primary tumor. We report a patient in whom the origin and the growth of HCC, together with the site of recurrence were exclusively endoluminal, without any continuity with a visible HCC in the liver parenchyma.

A 63-year-old man was referred for jaundice, pruritus and progressive elevation of the cholestatic liver function tests. Medical history included inactive hepatitis B and C infection, which had been treated successfully by interferon seven years previously. The patient did not follow any surveillance program since then. Serum alanine aminotransaminase (117-U/L [normal: 14—50 U/L]), aspartate aminotransaminase (73-U/L [normal: 12—50 U/L]), alkaline phosphatase (208-U/L [normal: 30—125 U/L]), gamma-glutanyl-transferase (319-U/L [normal: 9—40 U/L]), and total bilirubin (227-μmol/L [normal: 7—25 μmol/L]) levels were elevated. The CA 19-9 marker was elevated (249-kU/L [normal: < 37 kU/L]), whereas carcinoembryonic antigen and alpha-fetoprotein levels were within the normal range.

Contrast-enhanced abdominal computerized tomography (CT) and liver magnetic resonance imaging (MRI) showed an obstruction of the biliary confluence extending to the left hepatic duct and marked upstream dilatation of intrahepatic bile ducts. The obstruction was caused by a focal, intraductal, solid lesion displaying hypointensity on both T1- and T2-weighted images, mild contrast enhancement on arterial phase images and subtle washout on portal venous and delayed phase images with regard to liver parenchyma (Fig. 1). There were no signs of periductal invasion of the

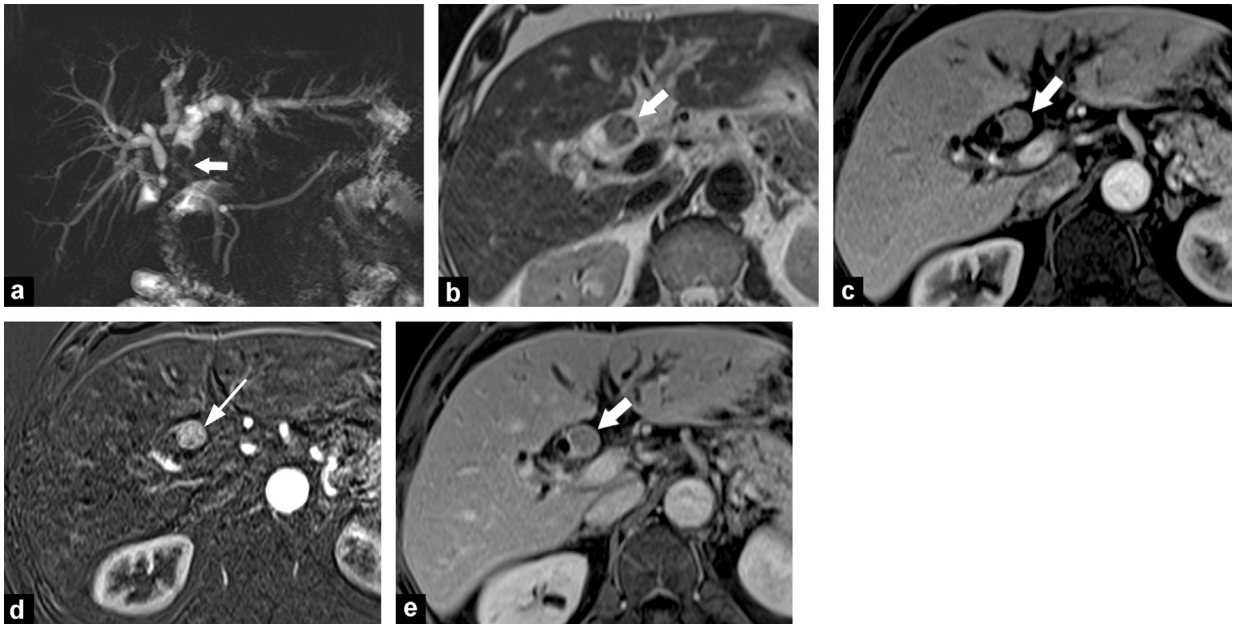

Figure 1. Preoperative MR imaging of the liver and biliary tract in a 63-year-old man with endobiliary hepatocellular carcinoma: a: MR cholangiography image in the coronal plane shows markedly hypointense endobiliary tumor (arrow) located at the biliary confluence; b: on T2-weighted fast spin echo MR image in the transverse plane, the tumor (arrow) is isointense relative to the hepatic parenchyma; c: on T1-weighted MR image obtained during the arterial phase after IV administration of a gadolinium chelate, the tumor (arrow) shows enhancement similar to that of hepatic parenchyma; d: endobiliary tumor enhancement (arrow) is more visible on subtracted image; e: delayed phase image shows subtle washout. No biliary wall thickening or sign of peribiliary invasion is visible on any sequence.

hepatic artery, the portal vein or the liver parenchyma. Percutaneous transhepatic cholangiography before biliary drainage confirmed the presence of an intraductal mass within the biliary confluence (Fig. 2). The patient underwent resection of the main bile duct and biliary confluence, with a Roux-en-Y bilio-enteric anastomosis. A remote 2.1-cm nodule, not visible on preoperative imaging, was discovered in segment II by manual palpation and further resected. Gross examination of resected specimen showed a 4.5 cm, well-delineated, greenish endobiliary mass which was adherent to the bile duct with a focal invasion of the biliary wall and peribiliary space (Fig. 3). Histopathological analysis of both lesions led to the diagnosis of moderately differentiated HCC. The liver parenchyma showed extensive fibrosis with mild necro-inflammatory activity, graded as A1F3 according to the METAVIR score. Incomplete R1 resection of the tumor was observed. Twenty-five months after resection, an 8-cm hypervascular mass developed in the lumen of the bilio-enteric anastomosis, without extension into the liver parenchyma (Figs. 4 and 5). The patient underwent repeat resection of the lesion after opening of the Roux-en-Y loop. Histopathological analysis confirmed HCC recurrence, with microscopic invasion of the anterior wall of the intestinal loop. The disease recurred again 8 months later, with an endoluminal mass in the bilio-enteric anastomosis obstructing the biliary tree. The patient underwent external beam radiation therapy (total dose, 30 Gy), followed by four cycles of gemcitabine and oxaliplatin (GEMOX). At 6-month follow-up, CT showed partial response with biliary decompression.

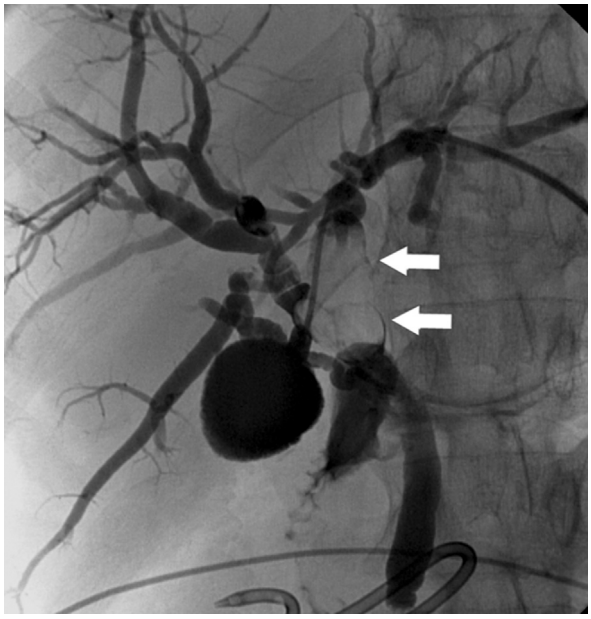

Figure 2. Percutaneous transhepatic cholangiography shows a well-defined and lobulated filling defect (arrows) at the biliary confluence, which extends to the left hepatic duct. The drain in projection of the bottom of the image was placed to evacuate a necrotic collection due to an acute pancreatitis complicating endoscopic retrograde cholangiography.

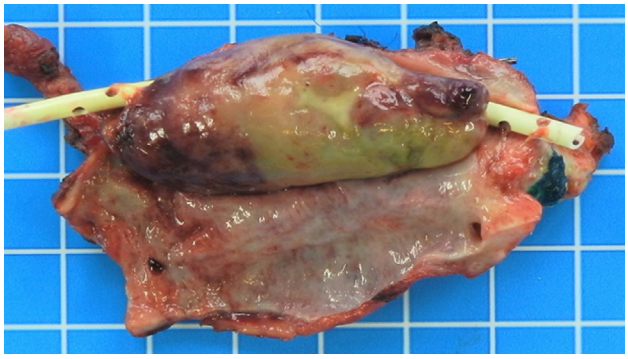

Figure 3. Surgical specimen presents as a greenish tumor mass with well-defined borders, which is adherent to the wall of the main bile duct. Note the presence of portion of biliary drain (yellow catheter).

The origin of this uncommon form of HCC remains unclear. Cho et al. hypothesized that it may develop directly from progenitor cells located in the biliary mucosa [3]. Another explanation is the presence of existing ectopic liver tissue, and possible endobiliary migration of distant lesion due to tumor hemorrhage or to a fragment of necrotic tumor separated from an invaded peripheral bile duct. Finally, the possibility of a primary tumor in the vicinity of the

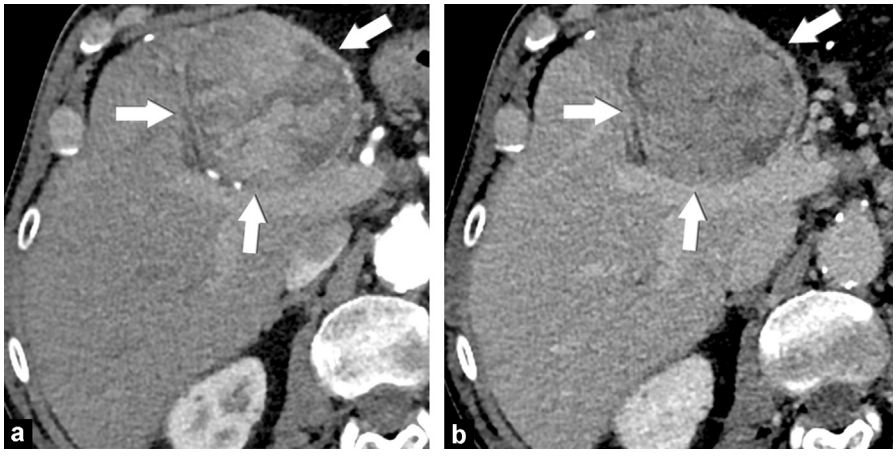

Figure 4. Recurrence of HCC at the bilio-enteric anastomosis: a: a lobulated and hypervascular mass (arrows) is visible at the bilio-enteric anastomosis on CT image obtained in the transverse plane during the arterial dominant phase; b: the mass (arrows) shows washout of contrast on delayed phase images.

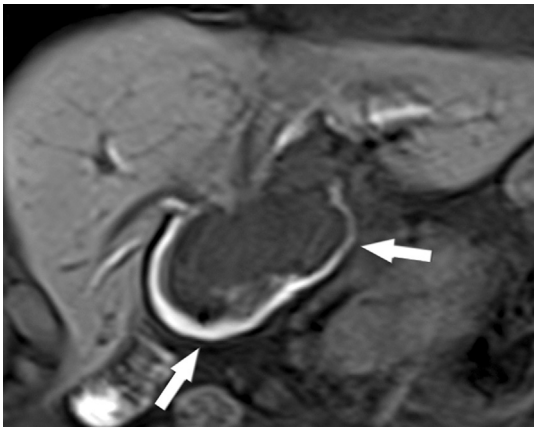

Figure 5. T1-weighted delayed image in the coronal plane after IV administration of gadoxetate disodium (Primovist®) shows hypointense endoluminal mass at the bilio-enteric anastomosis surrounded by the excreted contrast agent (arrows) which is hyperintense.

endobiliary HCC but too small to be identified cannot be excluded. Non-invasive diagnosis of this condition is very difficult, as most patients present with jaundice, and non-specific symptoms. As a consequence, most of similar cases are considered as cholangiocarcinomas, and HCC is an unexpected histopathological finding. However, the presence of an underlying chronic liver disease may help reach the proper diagnosis [4,5]. Imaging findings are not specific, but may be helpful, especially by showing enhancement of the intraluminal tumor in the biliary duct on the arterial phase [6]. Nevertheless, this feature is not specific for the diagnosis, and hypervascularity has also been described in rare forms of biliary tumors [7]. Indeed, the most important differentials are cholangiocarcinoma with endobiliary growth or rare liver intraductal papillary mucinous neoplasms of the liver. Metastatic disease may rarely cause biliary obstruction [8].

Treatment of endobiliary HCC is similar to that performed for primary biliary lesions. As in the present case, most patients in previous reports underwent bile duct resection with bilio-enteric anastomosis, associated with variable degrees of liver resection. In nearly all reported cases, the outcome was very poor, with early recurrence or death. The pattern of recurrence of the present case was very peculiar and reported only by Vibert et al. [9]. Indeed, the recurrent lesion also showed a mostly polypoid endoluminar growth in the bilio-enteric anastomosis, with limited perianastomotic invasion.

Disclosure of interest

The authors declare that they have no competing interest.

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