VIDEO CASE REPORT

A rare case of hepatocellular carcinoma with bile duct invasion diagnosed by peroral cholangioscopy

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CASE PRESENTATION

An 85-year-old man who had a history of hepatitis B infection and was being treated for prostate cancer presented with mild liver dysfunction. Laboratory examinations showed elevated levels of γ-glutamyl transpeptidase (365 IU/L) and alkaline phosphatase (727 IU/L), but transaminases and bilirubin were not elevated. For tumor markers, only carbohydrate antigen 19-9 was mildly elevated.

Abdominal ultrasonography revealed a 20-mm mass in the hilar region of the liver with internal heterogeneity and hypoechoic zones at the margins. Enhanced CT showed a relatively low-enhancing small tumor in the hilar bile duct and dilation of the left intrahepatic bile duct (Fig. 1). Endoscopic retrograde cholangiography and intraductal ultrasonography revealed a soft, movable, round tumor, measuring 10 mm in diameter (Figs. 2 and 3). Although these findings suggested hilar cholangiocarcinoma or intraductal papillary neoplasm of the bile duct, the specimens could not be adequately acquired by conventional biopsy during the first ERCP because of the movable nature of the tumor, and a diagnosis could not be made.

PROCEDURE

We decided to perform peroral cholangioscopy (POCS). A TJF-Q290V duodenoscope (Olympus, Tokyo, Japan) was inserted, and the POCS instrument (SpyGlass DS II, Boston Scientific, Natick, Mass, USA) was further inserted into the common bile duct. POCS showed a 20-mm-diameter, stalked, floating, round mass in the hilar region. The surface of the tumor was covered with white fur, and the tumor tissue was partially exposed in a map-like pattern. The mass was penetrable even with a guidewire, and it bled easily. Biopsies were performed using biopsy forceps,

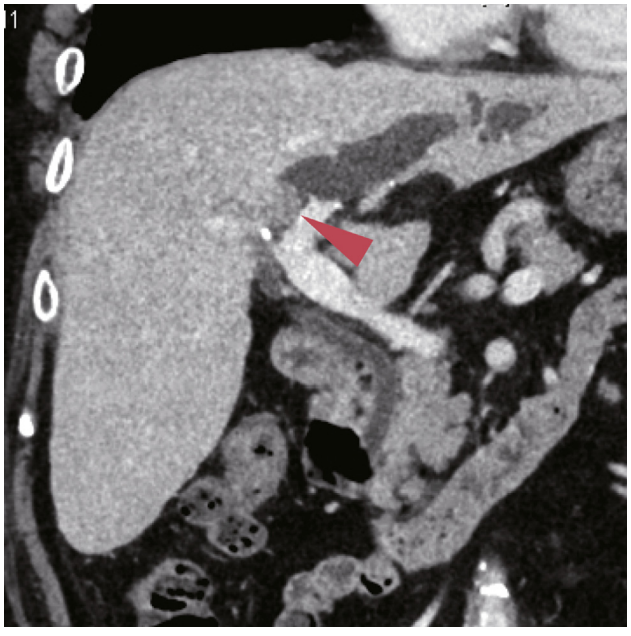

Figure 1. Contrast-enhanced CT showed an isoenhanced small tumor in the hilar bile duct and dilation of the left intrahepatic bile duct.

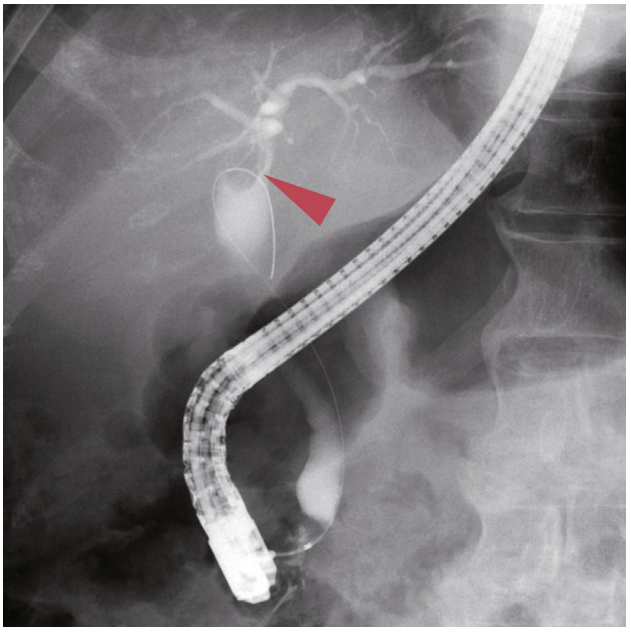

Figure 2. Endoscopic retrograde cholangiography showed a small round tumor in the hilar bile duct.

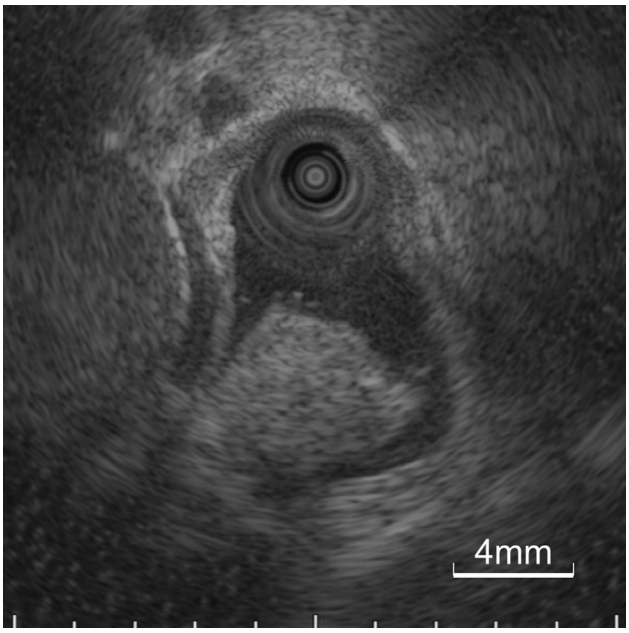

Figure 3. Intraductal ultrasonography revealed a movable, round tumor, measuring 10 mm in diameter.

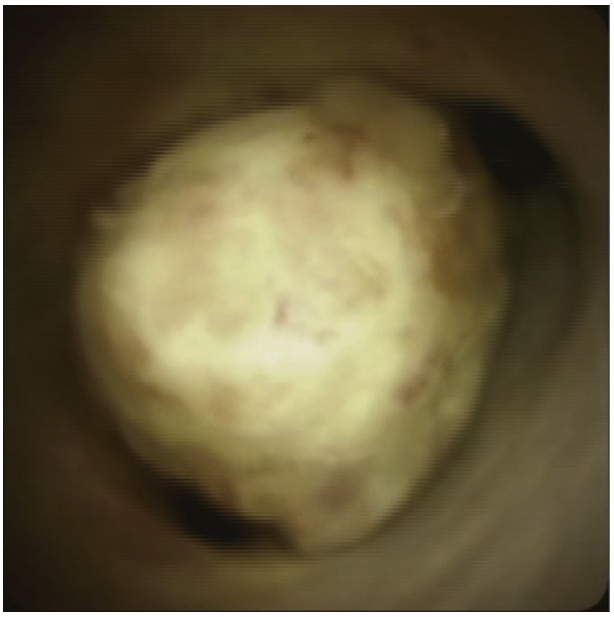

Figure 4. Peroral cholangioscopy revealed a 20-mm-diameter, stalked, floating, round mass in the hilar region. The surface of the tumor was covered with white fur, and the tumor tissue was partially exposed in a map-like pattern.

and the tumor bled easily even with biopsy (Fig. 4; Video 1, available online at www.giejournal.org).

OUTCOME

In the specimen, which was acquired by using POCS, the tumor cells had enlarged, round nuclei with clear or eosinophilic cytoplasm and presented in a cord-form structure. These cells were immunohistochemically positive for hepatocyte paraffin 1, but negative for cytokeratin 7 (Fig. 5). Surprisingly, these findings indicated that the tumor was hepatocellular carcinoma (HCC), which did not form tumors in the liver parenchyma but mainly formed masses in the bile ducts.

Although a diagnosis was made, the patient did not wish to undergo surgery or chemotherapy with lenvatinib, which we suggested, and preferred to be followed without treatment. Eight months later, the patient came to our hospital with cholangitis, and contrast-enhanced CT showed a mass that had grown to dilate the hilar bile ducts. Tumor invasion was identified in the liver that was contrasted in early phase and washed out in delayed phase (Fig. 6). ERCP was performed and stents were placed in the right and left hepatic ducts. The patient underwent palliative treatment and died 10 months after diagnosis.

DISCUSSION

HCC with bile duct invasion is reported to have a poorer prognosis than HCC without bile duct invasion.[1] Moreover, bile duct invasion of HCC, in which a primary tumor cannot be detected in the liver, is very rare.[2] Because of its low incidence and recognition, it is difficult to diagnose by imaging alone.[3] In a report of 9 cases of this type of HCC, no case could be diagnosed preoperatively, and almost all were diagnosed as intrahepatic cholangiocarcinoma.[4]

If tissue cannot be obtained by conventional biopsy, scraping of the mass is another option, although there is a risk of massive bleeding. The use of POCS has advanced diagnostic precision of biliary tract diseases,[5] which could lead to appropriate therapy. When a whitish, round, soft tumor is observed on POCS, as in this case, HCC should be included in the differential diagnosis, even if no tumor is evident in the liver.

DISCLOSURE

All authors disclosed no financial relationships.

Abbreviations: HCC, hepatocellular carcinoma; POCS, peroral cholangioscopy.

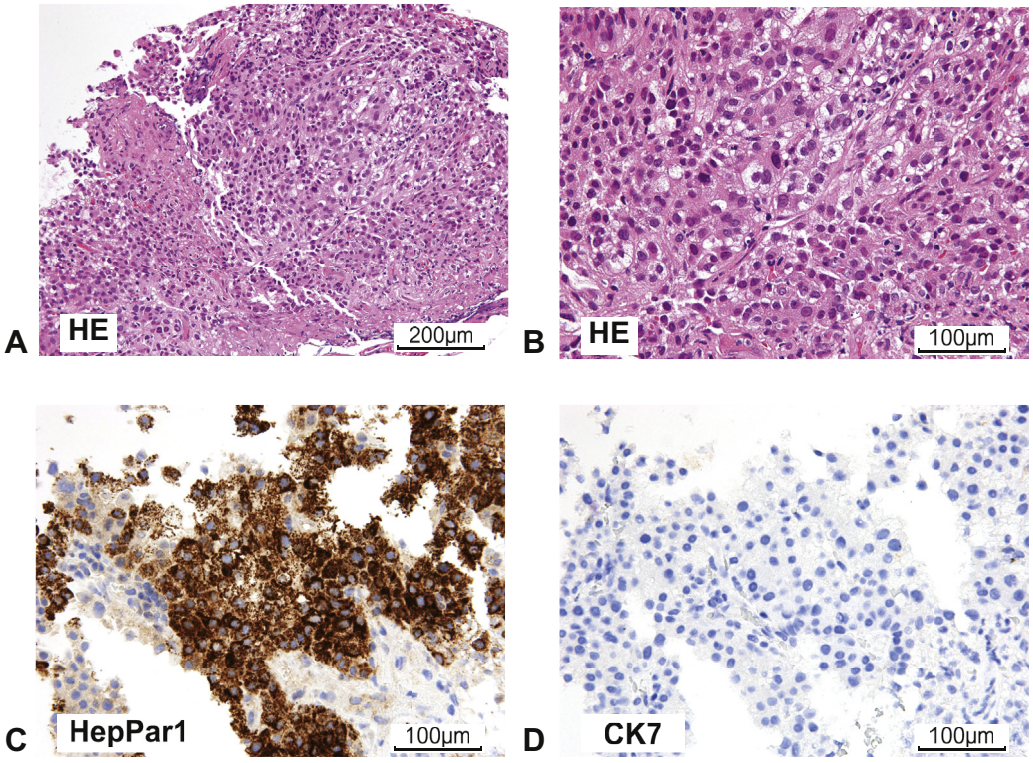

Figure 5. The tumor cells had enlarged, round nuclei with clear or eosinophilic cytoplasm and presented in a cord-form structure (hematoxylin and eosin stain) **(A, B)**. These cells were immunohistochemically positive for hepatocyte paraffin 1 **(C)** but negative for cytokeratin 7 **(D)**.

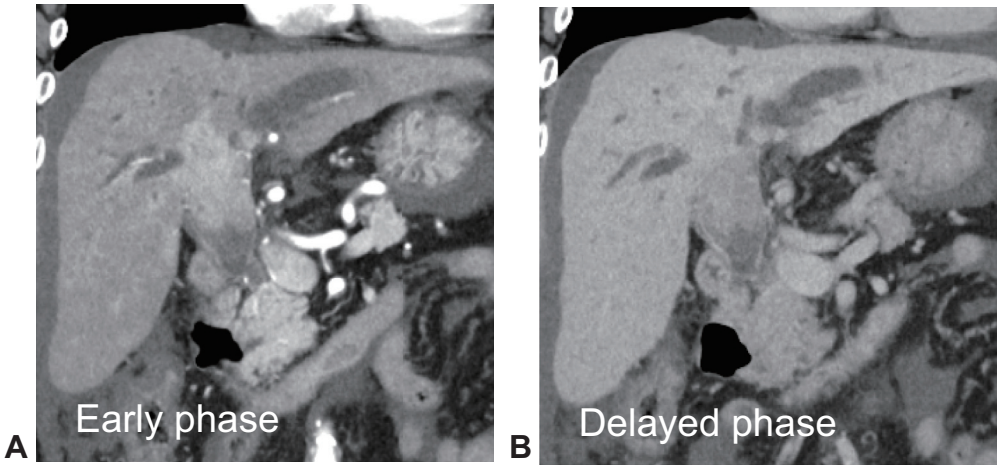

Figure 6. Dynamic CT 10 months after diagnosis. Contrast-enhanced CT revealed a mass that had grown to dilate the hilar bile ducts. Tumor invasion was identified in the liver that was contrasted in early phase **(A)** and washed out in delayed phase **(B)**.

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