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A case of cholangiolocellular carcinoma featuring intratumoral hepatic artery penetration: A case report

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ABSTRACT

INTRODUCTION: Cholangiolocellular carcinoma (CoCC) is thought to originate from hepatic stem cells. Its clinical characteristics, including radiological and prognostic factors, remain unclear.
PRESENTATION OF CASE: A 79-year-old woman with hypertension was admitted to our hospital after abnormal tumor marker levels were detected during an annual physical examination. Her laboratory data results were within normal range, and she was classified as Child-Pugh A. Enhanced computed tomography revealed a tumor located on the left side of the liver, with a maximum size of 60 mm. The tumor showed heterogeneously enhancing edges in the arterial phase, while prolonged tumor enhancement was detected in the delayed phase. Tumor penetration by the left hepatic artery was evident, whereas the left portal vein was invaded by the tumor. The preoperative diagnosis was cholangiocellular carcinoma. Left hepatectomy and cholecystectomy were performed with no postoperative complications; the final diagnosis was CoCC. Multiple liver metastases appeared 6 months after surgery; the patient is now receiving systematic chemotherapy.
DISCUSSION: While portal vein penetration into CoCCs has been reported, the same is not true of the hepatic artery; therefore, this case illustrates a unique tumor growth pattern.
CONCLUSION: A unique growth pattern as well as a large primary tumor may contribute to earlier recurrence.

1. Introduction

Cholangiolocellular carcinoma (CoCC) is a rare type of liver cancer [1,2], and was first reported by Steiner and Higginson in 1959 [1]. CoCC is derived from the Canals of Hering, or cholangioles, where hepatic progenitor cells (HPCs) are located [3]. HPCs have the potential to differentiate into hepatocytes and cholangiocytes [4], and expression of the HPC markers CD56, CD13, and epithelial cell adhesion molecule (EpCAM) are characteristic of CoCC [5].

It is difficult to diagnose CoCC preoperatively without histopathological anaysis; its radiological attributes are very similar to those of cholangiocellular carcinoma (CCC) and malignant lymphoma, which are characterized by tumor enhancement patterns and a specific shape on contrast-enhanced computed tomography (CT) [6]. Vascular penetration into the tumor, with no capsule present, is one of the notable characteristics of CoCC [7]. The disease has a post-curative surgery prognosis that lies between hepatocellular carcinoma (HCC) and CCC, in terms of favorable outcomes [8,9].

We present a patient with CoCC who underwent curative surgery, although multiple liver recurrences were observed 6 months later. We also conduct a literature review on CoCCs. This work has been reported in line with the SCARE criteria [10].

2. Presentation of case

The patient was a 79-year-old woman with hypertension; she had no relevant medical history and did not abuse alcohol. Elevated carbohydrate antigen 19-9 (CA19-9) was noted during an annual medical examination, and she was admitted to the Department of Surgery at our institution. Her physical examination was normal; however, her levels of tumor markers were as follows: CA19-9, 370 U/mL (normal, <37.0 U/mL); carcinoembryonic antigen, 1.6 ng/mL (normal, <5.5 ng/mL); alfa-fetoprotein, 2.2 ng/mL

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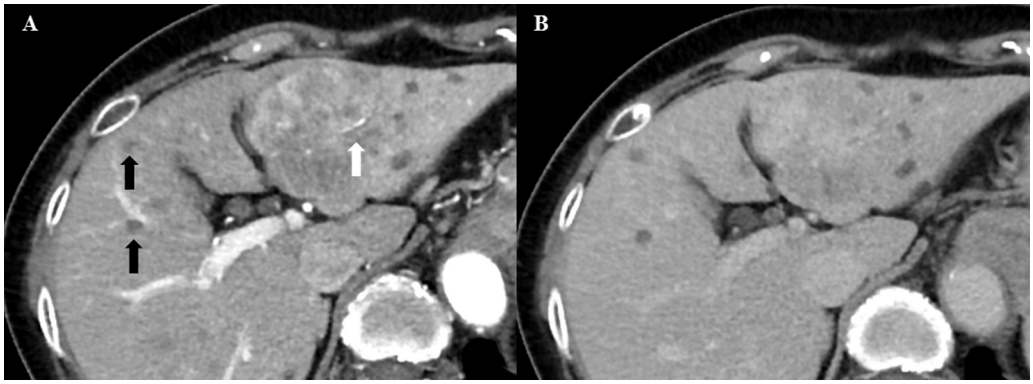

Fig. 1. Preoperative computed tomography. (A) The tumor in the left hepatic lobe showed enhancement from its periphery, with penetration of the hepatic artery and the dilatation of the peripheral biliary duct in the arterial phase (white arrow). Liver cysts were observed in segments 5 (black arrow). (B) Tumor enhancement was prolonged in the equilibrium phase.

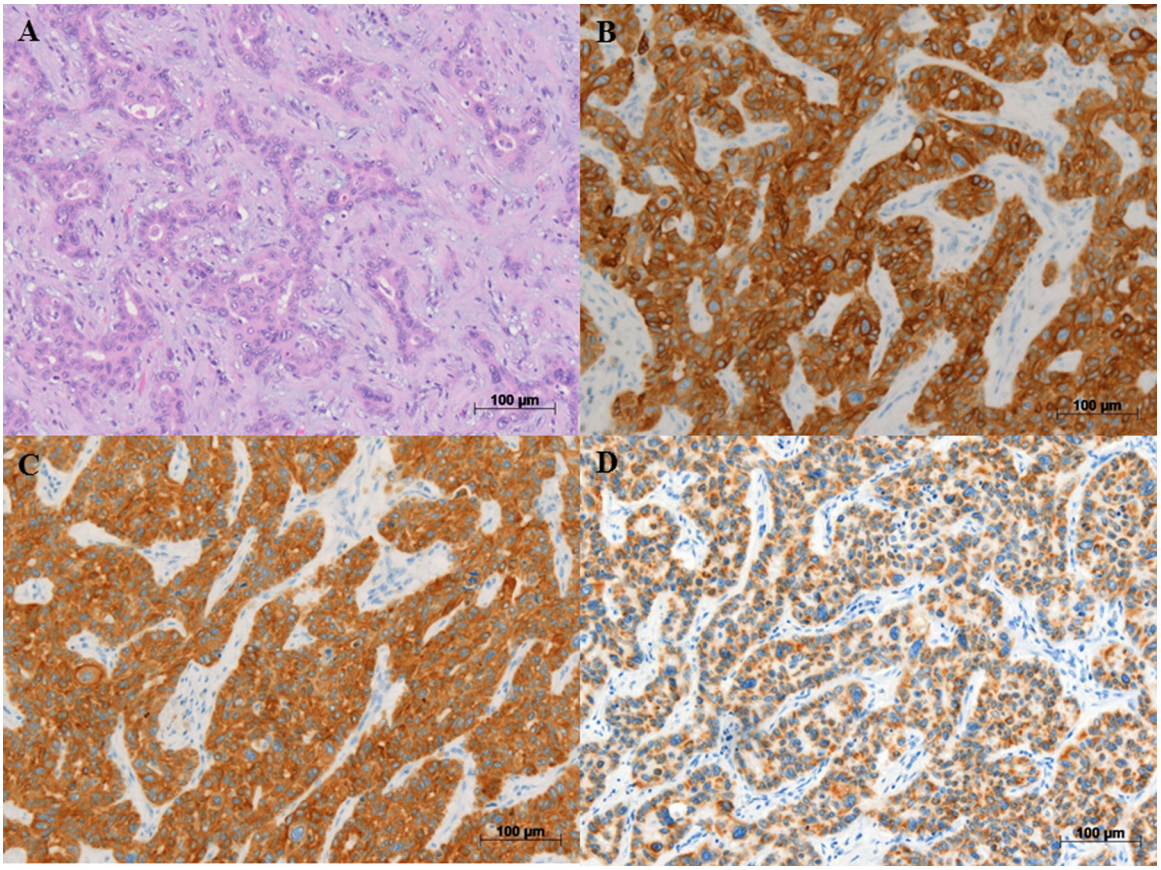

Fig. 2. Immunohistochemical findings of the tumor. (A) Hematoxylin and eosin staining: The tumor was composed of small glands showing antler-like and anastomosing patterns with abundant fibrous stroma. The tumor was positive for cytokeratin (CK) 7 (B), CK19 (C), and epithelial cell adhesion molecule (D).

(normal, <10.0 ng/mL); and protein induced vitamin K absence or antagonists-II, 20 mAU/mL (normal, <40 mAU/mL). Hepatitis virus markers were all negative. Her laboratory data indicated a Child-Pugh A classification. Contrast-enhanced CT revealed a tumor located in segment 4 measuring 60 mm (at most) with an unclear border. In early-phase imaging, the tumor showed enhancement at the tumor periphery; this was prolonged on late-phase imaging (Fig. 1). With magnetic resonance imaging (MRI), the tumor showed low and high intensities on T1-and T2-weighted images, respectively. This enhancement was washed out in the hepatocellular phase. Interestingly, the hepatic artery was observed penetrating the tumor on both CT and MRI. The patient's preoperative diagnosis was CCC following which left hepatectomy and cholecystectomy were performed. The duration of the surgery was 262 min, and the bleeding volume was 200 mL. Macroscopically, the tumor in segment 4 of the liver was a $60 \times 55 \times 50$ mm well-defined yellowish-white lesion. Histological examination revealed that the proliferating tumor cells replaced the surrounding normal tissue, and were composed of antler-like and anastomosing patterns with abundant fibrous stroma. There was no invasion into the vessels or lymph ducts. Immunohistochemical examination revealed positive expression of cytokeratin (CK) 7, CK19, and EpCAM (Fig. 2). Hence, the final diagnosis was CoCC.