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Case Report

Asymptomatic hepatobiliary cystadenoma of the hepatic caudate lobe: a case report

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Abstract

Human hepatobiliary cystadenoma is a rare benign cystic tumor of the liver, and is extremely rare in the caudate lobe. We herein present a case of a 70-year-old male with a hepatobiliary cystadenoma originating from the caudate lobe.

Keywords: liver, biliary tract, cystadenoma, MRI, ultrasonography

INTRODUCTION

Hepatobiliary cystadenoma has a very low incidence and is a multilocular cystic tumor of the liver. The majority of the cases are localized in the right side of the hepatic parenchyma[1]. Hepatobiliary cystadenoma originating from the caudate lobe is extremely rare and to date, only a small number of cases have been reported. Herein, we report a unique case of a 70-year-old male with hepatobiliary cystadenoma originating from the caudate lobe, who had no prior history of liver disease; the cystadenoma was incidentally found during a physical health examination.

CASE REPORT

A 70-year-old man was admitted to our hospital for physical health examination. The patient was in general good health and had no prior history of liver disease. He had never lived in pastoral areas and did not have a previous history of hepatic hydatid disease. Clinical examination was normal. When he underwent an abdominal ultrasound, a multilocular cyst measuring 6 cm×4 cm with internal septations was found in the caudate lobe of the liver (***Fig. 1***). In addition, sparse asterism-shaped blood flow signals were detected by color Doppler flow imaging. Before operation, the patient was re-evaluated with an abdominal MRI, which showed that the intrahepatic cystic part had uniform liquid signal. Contrast enhanced MRI showed that the wall septation and mural nodule were enhanced (***Fig. 2***). The patient was scheduled for surgery based on the ultrasound and MRI findings. Caudate lobectomy was performed to completely remove the lesion. Histopathological analysis yielded a diagnosis of typical hepatobiliary cystadenoma (***Fig. 3***). The epithelium showed strong staining with antibodies to CK7, CK8, CK19 and CK18, but was negative for AFP, CEA and

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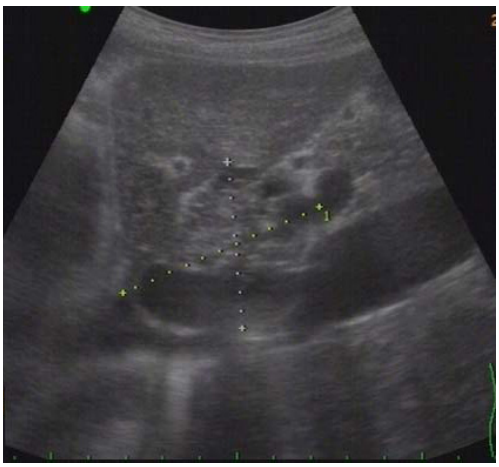

Fig. 1 **Sonogram showing a multilocular cyst with internal septations in the caudate lobe of the liver of a 70-year-old man**.

CK20. The proliferation index by Ki-67 staining was very low (< 5%). A final diagnosis of hepatobiliary cystadenoma was established. No evidence of recurrence was found during 6 months of follow-up.

DISCUSSION

Hepatobiliary cystadenoma is a rare tumor, and it may be found at any age. Approximately 85-95% of those affected are women, which suggests possible hormonal influence[4,5]. The incidence of this disease is very low in intrahepatic tumors, and about 200 cases have been reported throughout the literature since the first case of hepatobiliary cystadenoma was reported. They can lead to hepatomegaly, infection, bleeding, jaundice, and even obstruction in the vena cava[3].

Our case is interesting because hepatobiliary cystadenoma originating from the caudate lobe was found incidentally during a routine physical health examination. There are only two reported cases of hepatobiliary cystadenoma originating from the caudate lobe, and both cases were symptomatic[2,3]. It has been reported that hepatobiliary cystadenoma prolapses into the extrahepatic bile duct[6]. The clinical symptoms are atypical because of slow growth, so they are cur–

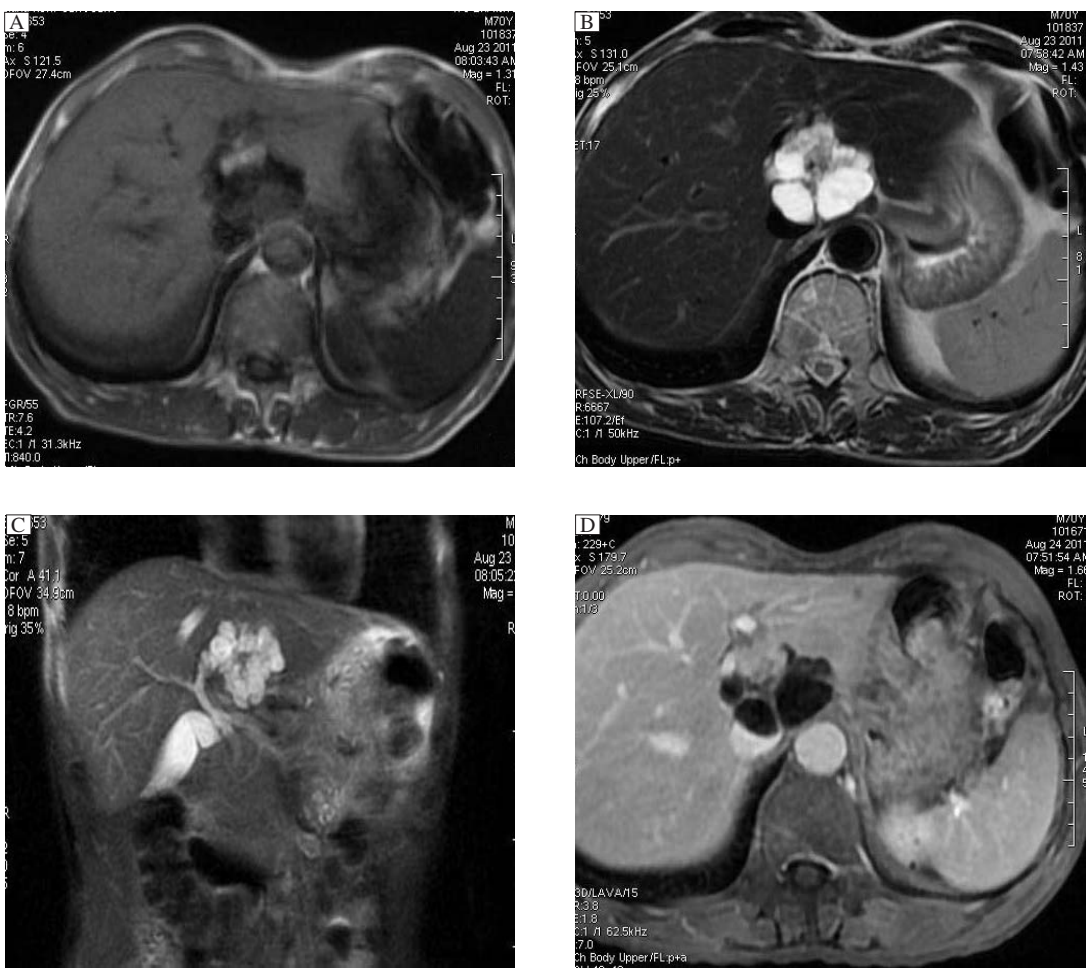

Fig. 2 **Magnetic resonance imaging of the abdomen.** The results show a low intensity area on T1WI (A), and a high intensity area on T2WI (B) as a multilocular cystic lesion with mural nodule, and petal-like high intensity area on 2D-FIESTA(C), and LAVA dynamic contrast-enhanced scan showing enhanced mural nodule and internal septa (D).

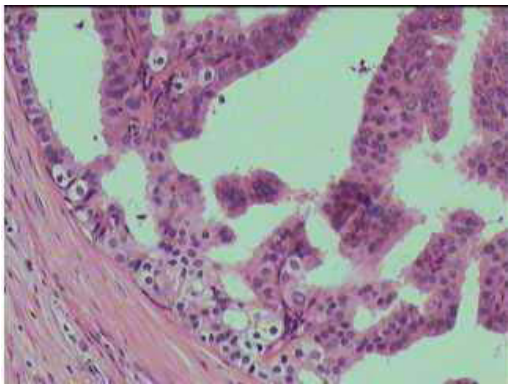

***Fig. 3* Pathological examination (H&E×100).** The result shows the cystic wall, most of which was lined with a single layer of columnar and cuboidal cells with no nuclear atypia.

rently found by imaging diagnostic techniques. Imaging studies by US, CT and MRI play a very important role in detecting the lesion. Ultrasonography could reveal a thick walled cystic mass, in which irregular septation can be demonstrated and also with perforator vessels in color Doppler flow imaging. MRI improves tissue characterization because of its high contrast resolution. On T1-weighted images, the signal intensity may change from hypointense to hyperintense as protein concentration increases. T2-weighted images demonstrate fluid collections within the tumour with homogeneous high signal intensity while the wall of the mass shows a low-signal-intensity rim[7]. It is reported that mural nodules were seen within biliary cystadenocarcinoma[7,8]. As shown in our case and previously reported cases, enhanced mural nodules can be found on the cyst wall[9]. However, due to the lack of typical imaging features, it may be very difficult to render a correct diagnosis. Zhang et al.[10] presented the iconographic representations of hemorrhagic simple hepatic cyst mimicking hepatobiliary cystadenoma and showed that the cystic wall was unevenly thickened and there were some flame-like prominences on the wall. If elevated CA19-9 and CEA are detected in the cystic fluid or the epithelial lining, it is helpful to the diagnosis of hepatobiliary cystadenoma[11]. Since it is believed to be premalignant, complete resection is the best treatment[6,12]; meanwhile, it is very important to follow up patients with hepatobiliary cystadenoma.

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