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Imaging presentation of biliary adenofibroma: A case report

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Abstract

BACKGROUND

Biliary adenofibroma (BF) is a rare benign epithelial tumor with the possibility of malignant transformation. Its main pathological feature is a well-defined cystic or honeycomb mass. BF has no specific clinical manifestations or laboratory and imaging findings; thus, it is easily misdiagnosed before surgery. This report describes a case in which biliary cystadenoma was misdiagnosed preoperatively and BF was diagnosed postoperatively. The imaging features, particularly the magnetic resonance imaging (MRI) features, were analyzed and summarized.

CASE SUMMARY

A 68-year-old Chinese man was admitted to our hospital with a 2-mo history of abdominal discomfort. Following admission to our hospital, laboratory examinations showed normal tumor marker concentrations and liver function. Hepato-cellular carcinoma was considered after contrast-enhanced ultrasound examination. MRI suggested the possibility of cystadenoma of the bile duct. However, postoperative pathological examination confirmed the diagnosis of BF. No local recurrence was found 1 mo after surgery.

CONCLUSION

Our objective is to highlight the imaging diagnostic value of BF, especially on an MRI enhanced scan with gadolinium ethoxybenzyl diethylenetriamine pentaacetic acid.

Key Words: Biliary adenofibroma; Magnetic resonance imaging; Gadolinium ethoxybenzyl diethylenetriamine pentaacetic acid; Misdiagnosis; Case report

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Core Tip: The imaging presentation of biliary adenofibroma is complex and diverse, and the clinical history and laboratory examination were not specific. In this case, magnetic resonance imaging characteristics of biliary adenofibroma, especially enhanced scan with Gd-EOB-DTPA and an intravoxel incoherent motion diffusion-weighted imaging sequence were valuable.

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INTRODUCTION

In 1993, Tsui *et al*[1] were the first to describe biliary adenofibroma (BF), which is a benign, complex, tubulocystic liver tumor with a bland spindle-cell stromal component. BF is a rare disease that is usually misdiagnosed due to its nonspecific clinical symptoms and laboratory examination findings. Imaging [including ultrasound, computed tomography (CT), and magnetic resonance imaging (MRI)] usually reveal a multicystic tumor or complex mass with both solid and cystic components; these findings are very similar to cystadenoma or cystadenocarcinoma[2], intrahepatic cholangiocarcinoma[3], liver metastasis[4], and other lesions. However, the MRI findings have certain characteristics. We herein describe a case in which we analyzed the contrast-enhanced ultrasound and MRI findings of a patient with BF who was misdiagnosed with hepatocellular carcinoma by contrast-enhanced ultrasound and with hepatic cystadenoma by MRI before surgical resection of the lesion. We also summarize the imaging findings of BF in previous literature.

CASE PRESENTATION

Chief complaints

A 68-year-old man reported abdominal discomfort without obvious inducement for 2 mo.

History of present illness

The patient had no other symptoms.

History of past illness

His past medical history indicated hypertension and mild cerebral infarction for more than ten years. After regular drug treatment, these conditions were well controlled.

Personal and family history

He had no personal or family history of other diseases.

Physical examination

On physical examination, there was no tenderness or rebound pain in the abdomen, and percussion pain in the liver area was negative. His blood pressure was 121/75 mmHg, pulse rate was 67 bpm and body temperature was 36.4 °C.

Laboratory examinations

No abnormal laboratory examinations were observed, including liver function, tumor markers, infection markers, coagulation tests and complete blood count.

Imaging examinations

A plain CT scan at a local hospital showed a space occupying lesion in the left lobe of the liver and the patient was admitted to our hospital for further treatment. Ultrasonography showed hyperechoic nodules in the left lobe of the liver. Following injection of contrast agent, early and obvious enhancement was observed (Figure 1A). Plain MRI

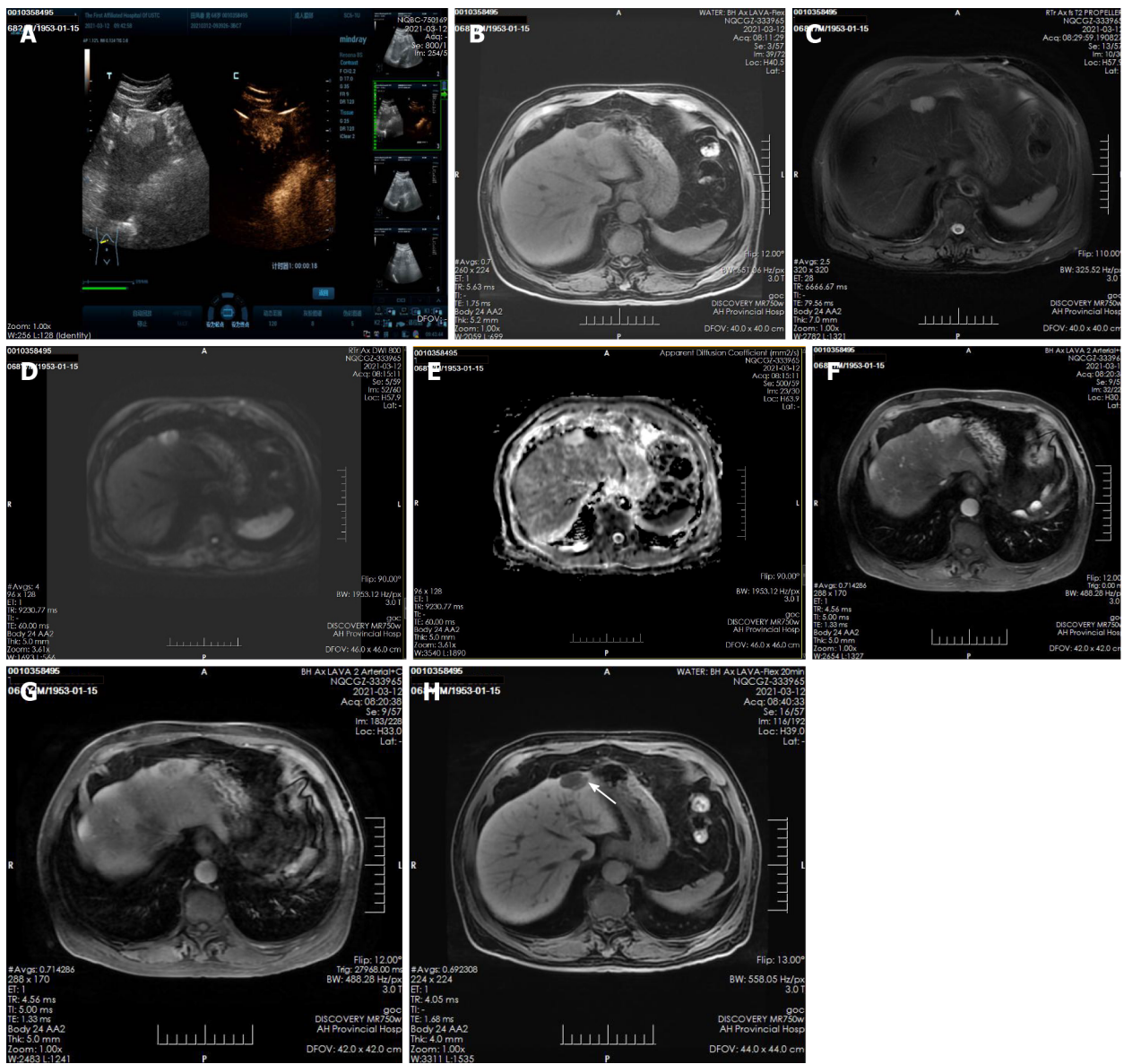


Figure 1 Imaging findings in a 68-year-old man with biliary adenofibroma. A: Ultrasonography showed hyperechoic nodules in the left lobe of the liver, after injection of contrast agent, it showed early and obvious enhancement; B and C: Plain magnetic resonance imaging (MRI) scan showed hypointensity on T1-weighted imaging (T1WI) and hyperintensity on T2-weighted imaging (T2WI); D and E: Diffusion-weighted imaging (DWI) showed moderate hyperintensity and isointensity on ADC; F-H: Enhanced MRI scan showed obvious enhancement of solid components and separation of lesions in the arterial phase, the degree of lesion enhancement decreased in the portal phase, in the hepatobiliary phase, enhancement of the bile duct structure was found in the lesion.

showed hypointensity on T1-weighted imaging (T1WI) and hyperintensity on T2-weighted imaging (T2WI). In this case, iterative decomposition of water and fat with echo asymmetry and least square estimation-iron quantification (IDEAL-IQ) and intravoxel incoherent motion diffusion-weighted imaging (IVIM-DWI) sequences were performed. The water phase of the IDEAL-IQ sequence showed that the signal of the lesion was higher than that of the liver. DWI showed moderate hyperintensity and isointensity according to the apparent diffusion coefficient (ADC). The ADC and pure diffusion coefficient (D) values were 2.78 and $2.12 \times 10^{-3} \text{ mm}^2/\text{s}$, respectively, which indicated that there was no obvious limitation in the diffusion of the lesion. There was no significant change in the signal of the lesion in the in-phase and out-phase sequence. The early and late arterial phases showed obvious enhancement of solid components and separation of lesions. The degree of lesion enhancement decreased in the portal phase and delayed scan. In the hepatobiliary phase, enhancement of the bile duct structure was found in the lesion (Figure 1B-H).

FINAL DIAGNOSIS

Pathology confirmed BF (a rare biliary epithelial tumor) in this patient (Figure 2). Microscopy showed irregular hyperplasia of the bile duct with varying amounts of intervening fibrous stroma and inflammatory cell infiltration. Some bile ducts were dilated and the wall thickened, cholestasis was seen in the bile duct. The cells showed no atypia, and some of them showed apocrine secretion.

TREATMENT

The lesion was removed by laparoscopic surgery under general anesthesia.

OUTCOME AND FOLLOW-UP

The patient had no signs of recurrence at the 1-mo postoperative re-examination (Figure 3).

DISCUSSION

Currently, although BFs are classified as benign bile duct tumors and precursors by the 2019 World Health Organization tumor classification system[5], some cases of BF are characterized by malignant transformation, invasion, and even distant metastasis (Tsui *et al*[1] first proposed in 1993, postoperative pathology showed malignant transformation in 3 cases[6-8], 3 cases of biliary adenofibroma with invasive carcinoma [9-11], 3 cases were complicated with liver tissue invasion[12-14], 2 cases were complicated with lymph node metastasis[15], local recurrence occurred in 2 cases[16]). The clinical manifestations and imaging characteristics of BF are nonspecific. A definitive diagnosis is difficult to achieve by preoperative imaging. The diagnosis mainly depends on postoperative pathological examination. Pathologically, the lesions are gray or dark red irregular masses that can show cystic, honeycomb, or solid changes and have a relatively clear boundary and no capsule.

Including the present case, 25 cases of BF have been reported to date (Tsui *et al*[1] in 1993, Parada *et al*[17] in 1997, Haberal *et al*[12] in 2001, Akin *et al*[18] and Garduño-López *et al*[19] in 2002, Gurrera *et al*[20] in 2010, Kai *et al*[6] and Nguyen *et al*[7] in 2012, Jacobs *et al*[13] in 2015, Godambe *et al*[9]; Thai *et al*[10] and Thompson *et al*[15] in 2016, Kaminsky *et al*[8] and Arnason *et al*[16] in 2017, Esteban *et al*[11] in 2018, Sturm *et al*[14] in 2019), and no differences in sex or age at onset are evident. The mean age at onset is 57.2 ± 17.79 years. Of the 25 cases reported 12 were male and 13 were female. The mean diameter of the lesions was 8.22 ± 4.65 cm. Eight patients presented due to upper abdominal discomfort and pain, one patient had jaundice due to an intrahepatic bile duct lesion, and the remaining 16 patients had no reported abdominal pain and were diagnosed by related examinations. The results of laboratory tests were generally nonspecific. One patient had an elevated carbohydrate antigen 19-9 (CA19-9) concentration[19], and all of the remaining patients had normal concentrations of tumor markers including alpha fetoprotein, carcinoembryonic antigen, CA19-9, and CA125. Liver function indices were within normal limits. One patient had a history of hepatitis B[21], but the hepatitis status was not mentioned in the remaining patients. The location of the lesion was the left lobe in 11 patients and the right lobe in 13 patients (the specific location of the liver lesion was not mentioned in the remaining patient). One patient had three lesions[16], and the remaining patients had single lesions. We carefully reviewed the previous literature (including the imaging findings and intraoperative descriptions of the lesion locations) and found that the most common location of BF was under the liver capsule, as was true in the present case. Thus, the lesion location has a certain particularity.

On plain CT and MRI scans, BF is mainly a cystic lesion containing septal and varied solid components. Only three lesions were described as solid tumors. In the present case, the lesion was small and mainly composed of solid components, and ultrasound showed that it was slightly hyperechoic. Plain MRI showed hypointensity on T1WI and hyperintensity on T2WI. In this case, IDEAL-IQ and IVIM-DWI sequences were performed. The water phase of the IDEAL-IQ sequence showed that the signal of the lesion was higher than that of the liver. DWI showed moderate

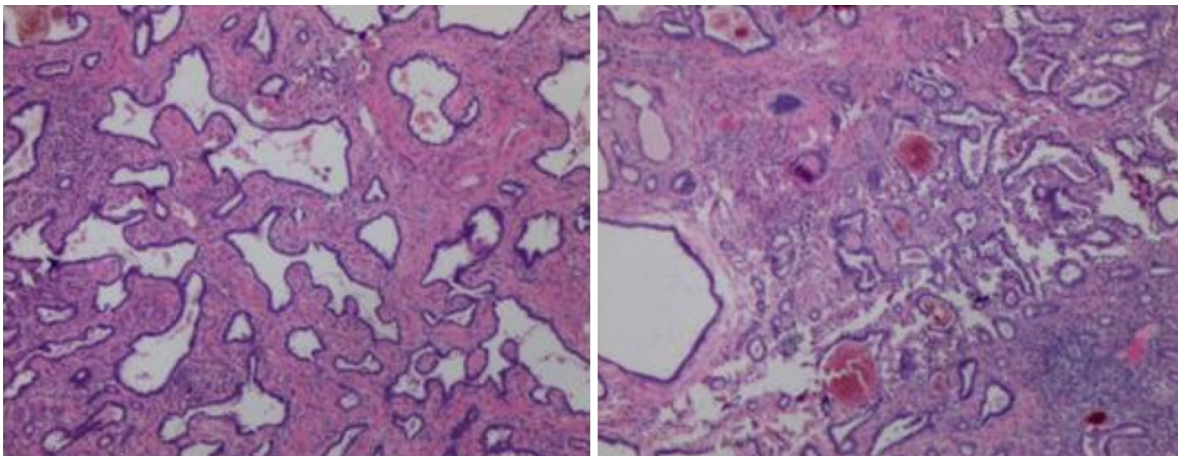


Figure 2 Postoperative pathology. Hematoxylin and eosin staining (100 ×) microscopy showed irregular hyperplasia of the bile duct with varying amounts of intervening fibrous stroma and inflammatory cell infiltration. Some bile ducts were dilated and the wall thickened, cholestasis can be seen in the bile duct. The cells showed no atypia, and some of them showed apocrine secretion. These results were consistent with the presentation of biliary adenofibroma.

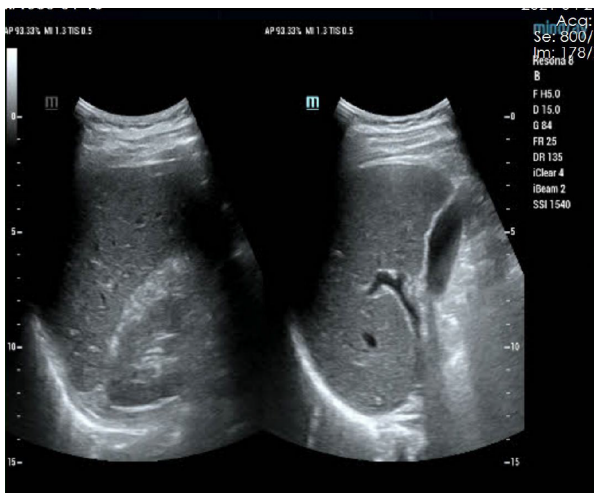


Figure 3 Postoperative re-examination images. Ultrasound examination indicates uniform echogenicity of liver parenchyma with no obvious abnormal echogenicity

hyperintensity and isointensity according to the ADC. The ADC and D values were 2.78 and $2.12 \times 10^{-3} \text{ mm}^2/\text{s}$, respectively, which indicated that it was more likely a benign lesion (there was no obvious limitation in the diffusion of the lesion). There was no significant change in the signal of the lesion in the in-phase and out-phase sequence. In previous cases, the bile duct epithelial components of the lesions had no secretory function, but some lesions showed a tendency to enlarge and develop cystic components during the follow-up period[21]. The diameter of the solid lesions in three cases[8,10,20] and the solid lesions in the present case were smaller ($4.08 \pm 1.16 \text{ cm}$) than the mean diameter of the cystic and solid lesions ($7.00 \pm 3.03 \text{ cm}$). Therefore, whether the bile duct epithelial components in BF have a secretory function requires further study. The intrahepatic bile duct was dilated in only one case (the lesion was located within the bile duct). Enhanced CT and MRI scans showed enhancement of the solid components and septa of the lesions; one report described a wash-in and wash-out enhancement pattern in the literature[8]. Gd-EOB-DTPA (Primovist, Bayer Schering, Pharma AG, Berlin), a hepatocyte-specific contrast agent, was used in this case. The early and late arterial phases showed obvious enhancement of solid components and separation of lesions. Areas of abnormal perfusion could be seen around the lesion. The degree of lesion enhancement decreased in the portal phase and delayed scan, and the areas of abnormal perfusion disappeared. A similar enhancement pattern was seen on contrast-enhanced ultrasound. In one case, the lesion displaced the hepatic vein[14], and in another case, the lesion was accompanied by adjacent hepatic tissue invasion[12], indicating malignancy. In two cases, enlarged

lymph nodes were found in the hepatic hilum, and postoperative pathological examination showed metastasis[15]. The remaining reports did not mention imaging signs of a malignant tumor, although pathological examination after surgical resection of the lesions showed that the lesions were combined with cytological atypia or invasive cancer and even exhibited distant metastasis.

Pathological examination revealed that the fibrous tissue matrix of the tumor showed the histological pattern of a partially cystic bile duct. The bile pigment component in the tumor duct indicated direct continuity between the lesion and the biliary system, although postoperative pathology showed that the lesion was not clearly connected with the bile duct[6,21]. Primovist is a hepatobiliary-specific contrast agent that can show the biliary system in the hepatobiliary phase. In this case, enhancement of the bile duct structure was found in the lesion in the hepatobiliary phase, suggesting that the lesion was closely related to the intrahepatic bile duct system; postoperative pathology also showed a bile duct structure and intrabiliary cholestasis. In a previous report, MRI failed to show a relationship between biliary duct adenofibroma and the intrahepatic bile duct. The author speculated that because most of the lesions were located under the liver capsule and the terminal bile duct was slender, it was difficult to show the relationship between the lesion and the bile duct by conventional MRI scanning sequences. Primovist-enhanced MRI, especially hepatobiliary phase images, was a good supplement.

CONCLUSION

BF is a rare biliary tumor that exhibits malignant transformation and usually occurs under the liver capsule. The lesions are mostly cystic and solid. On enhanced scans, the solid part can show a wash-in and wash-out enhancement pattern, and enhancement can be seen in the separation of lesions. The hepatobiliary phase of Primovist-enhanced MRI can better show the lesions and intrahepatic bile duct structure, providing more clues and value for the diagnosis. BF can be treated by surgical resection, but regular postoperative review is needed to identify recurrence or distant metastasis, such as that to the lung.

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