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CASE REPORT

A case of hepatic angiomyolipoma difficult to distinguish from hepatocellular carcinoma

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Abstract

We report a case of hepatic angiomyolipoma with uncommon clinical features. A 56-year-old man presented with a hepatic tumor in the caudate lobe. The tumor was hypoechoic on ultrasonography, showed early-phase hyperattenuation on enhanced computed tomography and did not absorb iron on superparamagnetic iron oxide-enhanced magnetic resonance imaging. Hepatocellular carcinoma was highly suspected, and the patient underwent hepatic resection. Histologically, the tumor was mainly composed of smooth muscle cells and contained small amounts of adipose cells and blood vessels. On immunohistochemical staining, the smooth muscle cells were positive for a melanocytic cell-specific monoclonal antibody. In cases with uncommon features of angiomyolipoma, it is quite difficult to distinguish angiomyolipoma from hepatocellular carcinoma.

Key words: Adipose cell; Hepatic angiomyolipoma; Hepatocellular carcinoma; HMB-45; Smooth muscle cell

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INTRODUCTION

Hepatic angiomyolipoma (HAML) was first reported by Ishak[1]. It is a rare benign tumor which is composed of a heterogeneous mixture of adipose cells, smooth muscle cells and vessels, and can be treated conservatively if spontaneous hemorrhage or malignant change does not occur[2-4]. The radiological features of HAML depend on the relative proportions of adipose cells[5]. For this reason, preoperative diagnosis of HAML is occasionally difficult, and it is not easy to differentiate from hepatocellular carcinoma (HCC). Herein, we report a case of HAML which was difficult to differentiate from HCC.

CASE REPORT

A 56-year-old Japanese man was admitted to our hospital for further examination of a liver tumor in the caudate lobe. He had no history of liver disease or hepatitis and did not drink heavily. Hepatitis B surface antigen and anti-hepatitis C antibody were negative. Serum levels of transaminase, α-fetoprotein and des-γ-carboxy prothrombin were within the normal range. On ultrasonography, the tumor was hypoechoic (Figure 1A). Enhanced computed tomography (CT) showed a hepatic mass with early-phase hyperattenuation and late-phase hypoattenuation, measuring 4.2 cm × 4.0 cm in the caudate lobe (Figure 1B and C). Magnetic resonance imaging (MRI) revealed hypointensity on T1-weighted images, hyperintensity on T2-weighted images and hyperintensity on diffusion weighted images (Figure 1D-F). The tumor did not absorb iron on superparamagnetic iron oxide-enhanced (SPIO) MRI

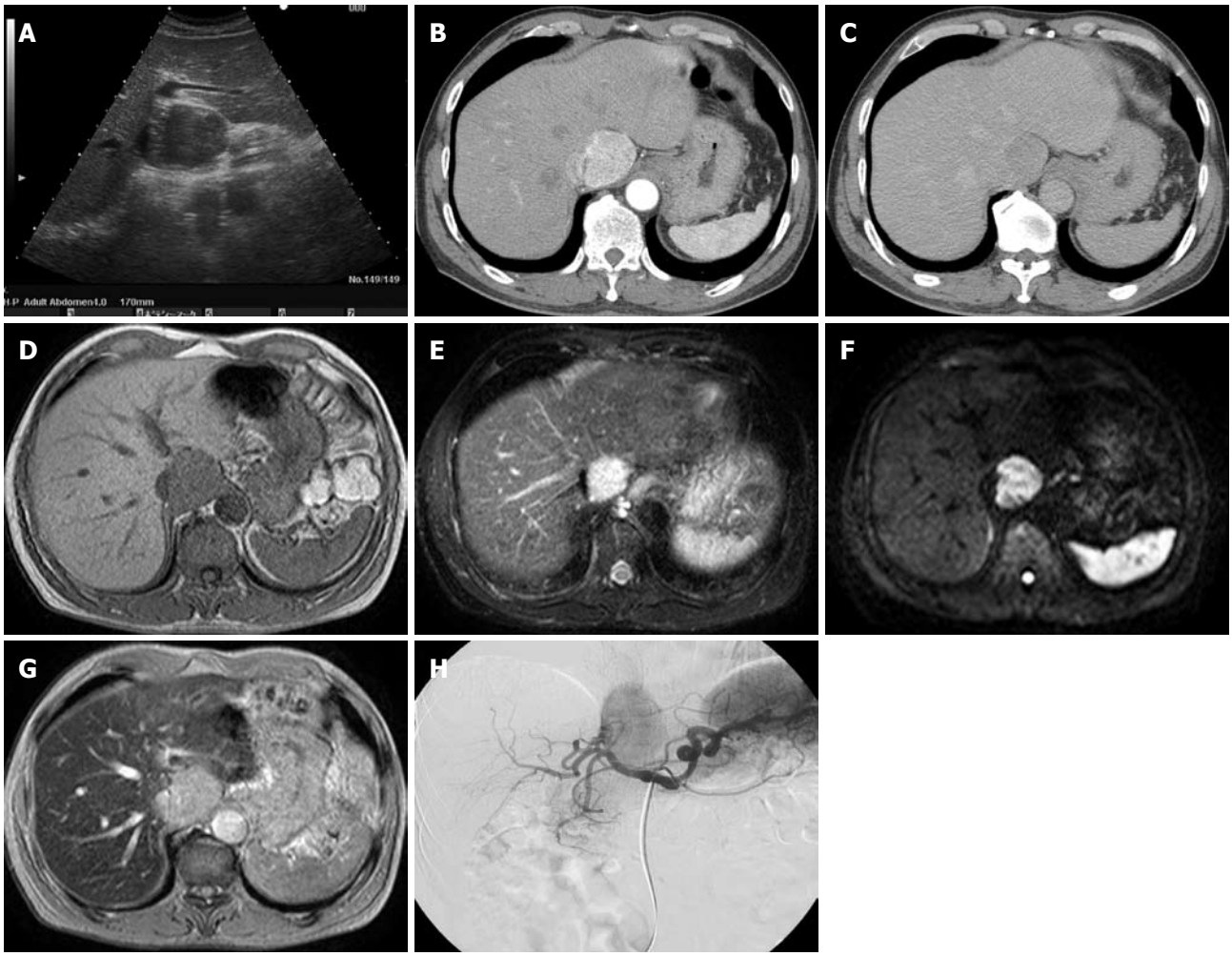

Figure 1 Images. A: The tumor was hypoechoic on ultrasonography, measuring 4.2 cm × 4.0 cm; B-C: Enhanced computed tomography showed a tumor with early-phase hyperattenuation and late-phase hypoattenuation; D-F: Magnetic resonance imaging showed a tumor with hypointensity on T1-weighted, hyperintensity on T2-weighted images and hyperintensity on diffusion weighted images; G: The tumor did not absorb iron on superparamagnetic iron oxide-enhanced MRI; H: On angiography, the tumor was shown as a circumscribed hypervascular mass.

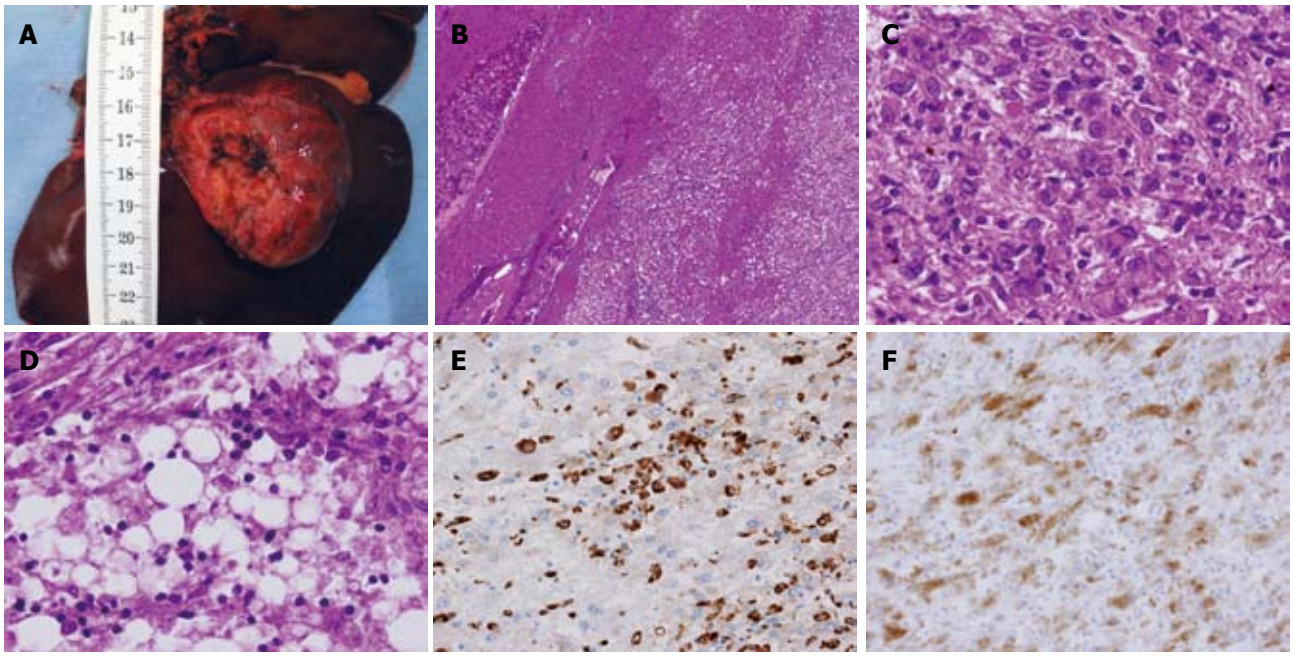

Figure 2 HAML. A: The tumor occupied a large area of the caudate lobe; B-D: The histological features of the tumor showed that it was mainly composed of smooth muscle cells (B: HE stain, × 4; C: HE stain, × 40) and a small number of adipose cells (D: HE stain, × 40); E, F: Immunohistochemically, the tumor was positive for CD68 (E: CD68, × 20), and HMB-45 (F: HMB-45, × 100).

(Figure 1G). On angiography, the tumor was shown as a circumscribed hypervascular mass (Figure 1H). HCC was highly suspected on radiological imaging. Resection of the left lobe with the tumor in the caudate lobe was performed. The tumor measured 6.0 cm in diameter, and the surface of the tumor was gray and white. Histologically, the tumor was mainly composed of smooth muscle cells and contained small amounts of adipose cells and blood vessels (Figure 2A-D). On immunohistochemical staining, the tumor was negative for desmin and S-100, but positive for actin and CD68, and the smooth muscle cells were positive for a melanocytic cell-specific monoclonal antibody (HMB-45) (Figure 2E and F). This tumor was diagnosed as HAML.

DISCUSSION

Recently, the concept of perivascular epithelioid cell tumor (PEComa) which was proposed by Bonetti et al in 1992 has gained wide acceptance[6]. PEComa is defined as a mesenchymal tumor composed of histologically and immunohistochemically distinctive perivascular epithelioid cells. Immunohistochemically, the tumors are consistently immunoreactive for HMB-45, a monoclonal antibody for melanoma. AML is considered a part of PEComa[7]. Smooth muscle cells of HAML stain positively for HMB-45. This finding is useful for the diagnosis of HAML, because liver tumors other than angiomyolipoma are negative for HMB-45[8].

In radiological diagnosis, HAML typically shows high echo on ultrasonography, early-phase hyperattenuation on enhanced computed tomography, hyperintensity on T2-weighted magnetic resonance imaging and a circumscribed hypervascular mass on angiography[9]. The soft tissue component (smooth muscle cells and vessels) is considered to be enhanced by the intravenous administration of contrast material[10]. However, the imaging features of HAML vary because of variations in the proportion of adipose cells, smooth muscle cells and vessels. In particular, the number of adipose cells varies between 10% and 90%[11]. HAML consisting of a small number of adipose cells shows low echo on ultrasonography and early-phase hyperattenuation on enhanced CT. These findings are similar to the imaging features of HCC[12]. On the other hand, there are no reports regarding the radiological features of SPIO MRI on HAML. SPIO contrast material, which is taken up by the reticuloendothelial system and depresses the signal of normal liver at T2-weighted imaging, is useful for the detection of hepatic tumors[13]. In our case, HAML was positive for CD68 stain, which is a Kupffer cell-related marker; however the signal at T2-weighted imaging on SPIO MRI was depressed similar to HCC. Thus, SPIO-MRI was not useful for differentiating HAML from HCC in our case.

HAML shows various histological patterns. According to the line of differentiation and the predominance of tissue components, the tumors are subcategorized into mixed, lipomatous ($\geqslant$ 70% fat), myomatous ($\leqslant$ 10% fat), and angiomatous types. The mixed type is the most common, but tumors with a small number of adipose cells such as the myomatous type, which are rare, show widely variable patterns in morphology[14]. In this study, we classified the HAML in our case as the myomatous type.

In conclusion, HAML is a benign tumor and requires no surgical treatment. However, the diagnosis is difficult because it has various histological patterns. In the myomatous type, the radiological findings including SPIO-MRO are similar to those of HCC. Hepatic tumors without obvious risk factors for HCC should be distinguished from HAML.

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