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A case of diffuse hepatic angiosarcoma diagnosed by FDG-PET

Ai Oe,* Daiki Habu,** Joji Kawabe,* Kenji Torii,* Etsushi Kawamura,* Jin Kotani,* Takehiro Hayashi,* Hiroki Sakaguchi** and Susumu Shiomi*

**Department of Nuclear Medicine and **Department of Hepatology, Graduate School of Medicine, Osaka City University*

A 76-year-old woman was admitted to our hospital with a 2-month history of increasing abdominal distension, leg edema, and dyspnea. The serum transaminase level was about twice the upper limit of normal. The CT showed no tumor. Fluorine-18 2-deoxy-2-fluoro-D-glucose (FDG)-positron emission tomography (PET) showed diffuse abnormal accumulation throughout the entire liver. She was diagnosed by histopathological examination as having hepatic angiosarcoma causing veno-occlusive disease (VOD). This is the first report of hepatic angiosarcoma with FDG-PET.

Key words: angiosarcoma, FDG-PET, VOD

INTRODUCTION

FDG-PET has attracted considerable attention recently in the field of oncology, because it can detect malignant tumors, distinguish between malignant and benign tumors, and evaluate tumor stage. FDG-PET can be used as a tool for follow-up study.[1,2]

We present a case of diffuse hepatic angiosarcoma. Conventional imaging methods (CT, MRI, and ultrasonography) did not show a malignant lesion. Using FDG-PET, we suspected a malignant liver tumor, and diagnosed angiosarcoma by histopathological examination.

CASE REPORT

A 76-year-old woman was admitted to our hospital with a 2-month history of increasing abdominal distension, leg edema, and dyspnea. A week before admission she visited the clinic, where she was found to have liver dysfunction, and was admitted to our hospital for further investigation.

On admission, she had massive ascites and a pleural effusion. Aspartate aminotransferase activity was 79 IU/*l*, alanine aminotransferase activity was 35 IU/*l*, alkaline phosphatase activity was 186 IU/*l*, total bilirubin concentration was 1.3 mg/d*l*, and albumin was 2.3 g/d*l*. The prothrombin time was 13.3 seconds. Serological tests were negative for hepatitis B and C. Antinuclear antibody was positive and her antimitochondrial antibody was negative. Alpha fetoprotein, carcinoembryonic antigen, and proteins induced by vitamin K antagonists or absence-II were within the normal ranges. The CT (Fig. 1) showed diffuse hypoattenuation, an irregular liver surface, and massive ascites, but no apparent mass lesion. FDG-PET (Fig. 2) showed an extensive diffuse abnormal accumulation throughout the entire liver, and the standardized uptake value (SUV) was 6.1. A HEAD-TOME IV SET-1400W-10 (Shimadzu Corp., Japan) was used for the PET study. We suspected malignant liver disease, and performed percutaneous biopsy under ultrasonographic guidance. Histopathological examination (Fig. 3) showed necrotic lesions around the central veins and many atypical cells which piled up into the central veins and hepatic sinusoids. Immunohistochemistry showed that the tumor cells were positive for CD34. Hepatic angiosarcoma and VOD were suspected. Her CT and FDG-PET showed no evidence of metastasis. We diagnosed hepatic angiosarcoma which had infiltrated the entire liver, causing VOD.

DISCUSSION

PET is now increasingly used in the field of oncology. PET can diagnose the quality of a tumor, while conventional imaging methods (CT, MRI, and ultrasonography)

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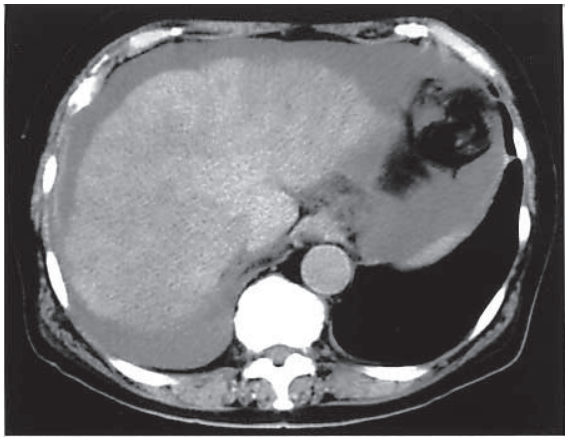
For reprint contact: Ai Oe, M.D., Department of Nuclear Medicine, Graduate School of Medicine, Osaka City University, 1–4–3 Asahimachi, Abeno-ku, Osaka 545–8585, JAPAN.
E-mail: oeai@med.osaka-cu.ac.jp



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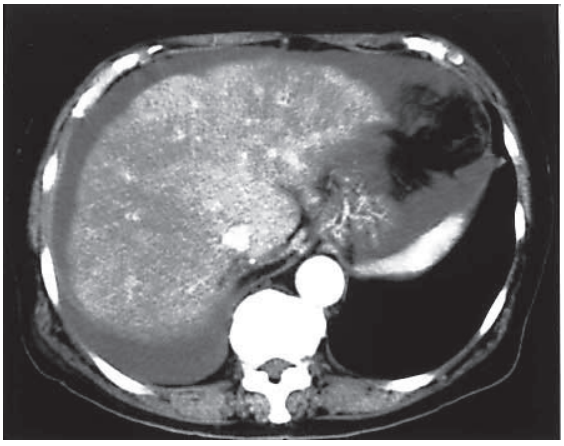

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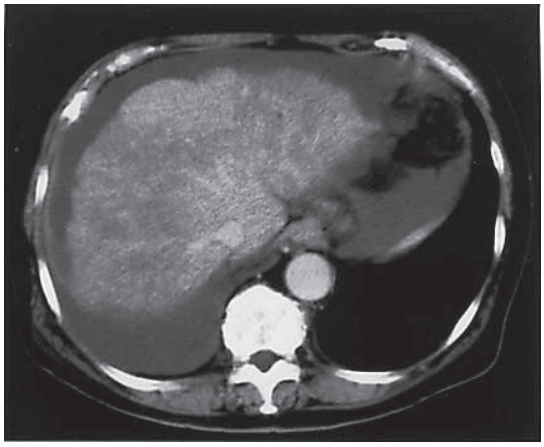

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Fig. 1 (a) Non-contrast CT scan shows diffuse hypoattenuation, an irregular liver surface, and massive ascites. Contrast CT scan in the early phase (b) and delay phase (c) shows no apparent mass lesion.

provide anatomical and morphological information. FDG is the most widely used tracer in oncology. The diagnosis of tumors with FDG-PET is based on increased regional glucose metabolism. The sensitivity of FDG-PET studies exceeds 85%, and is dependent upon tumor type, size, and location. Inflammatory lesions give false positives.[1]

Delbeke et al. assessed the value of FDG-PET in differentiating benign from malignant hepatic lesions.[2] They reported that metastatic liver tumors and cholangiocarcinomas have increased uptake values, with an SUV greater than 3.5. Hepatocellular carcinoma showed an increased FDG uptake or poor uptake. All benign hepatic lesions, including adenoma and focal nodular hyperplasia, had poor uptake, with an SUV of less than 3.5. At first we thought that this patient had liver dysfunction and ascites caused by liver cirrhosis. However, FDG-PET showed increased FDG uptake in the entire liver, with an SUV of 6.1. We therefore suspected a malignant tumor, and diagnosed diffuse infiltrating hepatic angiosarcoma by histopathological examination.

Angiosarcoma is a rare non-epithelial malignant tumor of vascular endothelial cells. This is a tumor which can arise in any organ—for example, skin, soft tissues, liver, spleen, bone, or breast. Primary hepatic angiosarcoma is very rare. Alrenga et al. reported that primary hepatic angiosarcoma constitutes about 1.8% of primary liver malignancies.[3]

The association of hepatic angiosarcoma with exposure to thorotrast, arsenic, vinyl chloride monomers,[4] anabolic hormones, and estrogens is well known, but many cases have no known cause; Locker et al. reported that 58% of cases have no known cause.[4] In this case there was no history of exposure to any carcinogens.

The prognosis is very poor. The median survival was reported to be 5.5 months by Locker et al.[4] and 6.9 months by Kojiro et al.[5] Some cases are complicated by multiple metastases because of rapid progression, DIC, intraperitoneal tumor rupture, the Kasabach-Merritt syndrome, or VOD. This case was complicated by liver dysfunction, pleural effusion, ascites, and edema caused by intrahepatic congestion due to VOD. We know of only one other reported case with VOD.[6]

Surgery can be a curative procedure, but most cases are unresectable at diagnosis, and only 20% of cases can be resected. Other therapies include transcatheter arterial embolization, chemotherapy (adriamycin or mitomycin), transhepatic arterial infusion chemotherapy with interleukin, radiation therapy, and liver transplantation. These therapies are not completely effective. This patient did

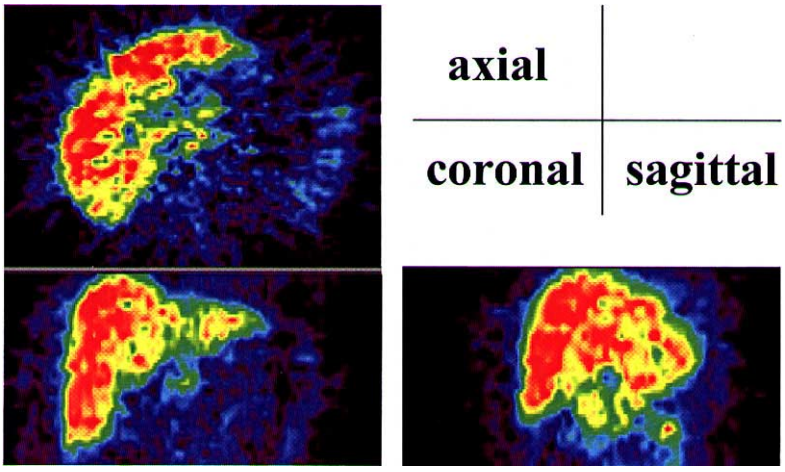

Fig. 2 FDG-PET shows extensive diffuse abnormal accumulation throughout the whole liver (SUV 6.1).

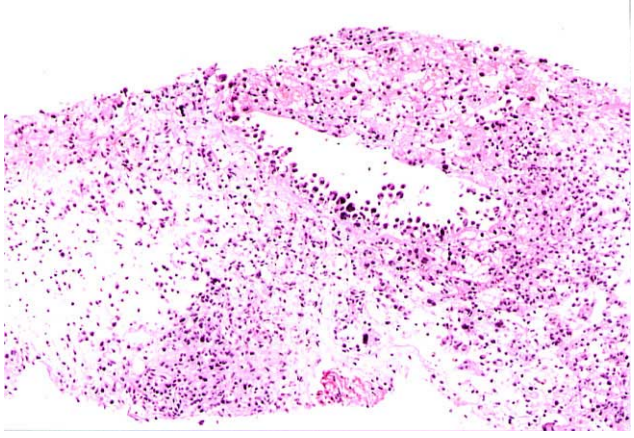

Fig. 3 Histopathological examination shows necrotic lesions around the central veins and many atypical cells which pile up into the central veins and hepatic sinusoids.

not receive active treatment because the tumor had diffusely infiltrated her liver, and her condition was poor.

Macroscopically, primary hepatic angiosarcoma is classified into multiple small nodules, a large dominant mass, large focal lesions, or diffuse infiltrating lesions.[7] In pathologic diagnosis, it is classified into sinusoid type and solid type. Factor VIII-related antigen, an endothelial cell marker, and CD31 are now used to aid in the diagnosis.[8] CT and MRI findings[7] are variable because there are many types of angiosarcoma. Angiography shows compression and extension of the hepatic artery, cotton wool-like pooling, a hypervascular area in the center of the tumor, and tumor stain at the end of hepatic artery. It is easy to differentiate it from HCC, but difficult to differentiate it from cavernous angioma.

To the best of our knowledge, there is no previous report of primary hepatic angiosarcoma in which PET was useful in diagnosis.

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