Radiat Med (2006) 24:369–372
DOI 10.1007/s11604-006-0034-1

CASE REPORT

Spontaneous rupture of a necrotic hepatic angiosarcoma: findings on dual-phase computed tomography and angiography

**Koshi Ikeda · Minoru Maehara · Naoto Ohmura
Hiroaki Kurokawa · Kenji Koda · Hidetaro Yokoyama
Satoshi Sawada**

Received: November 28, 2005 / Accepted: January 14, 2006

Abstract We present dual-phase computed tomographic (CT) and angiographic findings of a ruptured hepatic angiosarcoma. These tumors can be divided into two types: those with and those without gross central necrosis with hemorrhage. In our case, the tumor had gross central necrosis, and CT and angiographic findings showed a small number of areas with a centripetal enhancement pattern and the rest of the tumor with avascular areas. We found that dual-phase CT and angiographic findings are able to distinguish angiosarcoma, which mimics a hemangioma, as these lesions show avascular areas that reflect a mass with gross central necrosis.

Key words Liver · Angiosarcoma · Computed tomography · Angiography

Introduction

Hepatic angiosarcoma is a rare mesenchymal tumor that accounts for approximately 2% of all primary liver malignancies.[1] It is not uncommon for a hepatic angiosarcoma to present with spontaneous intraperitoneal hemorrhage. To the best of our knowledge, few reports have described the dual-phase computed tomography (CT) and abdominal angiography findings of a ruptured hepatic angiosarcoma.[2]

The CT appearance of angiosarcoma of the liver, although nonspecific, is consistent with a vascular tumor.[3] In a previous report, angiosarcoma was found to mimic cavernous angiosarcoma on dual-phase CT.[4] In our case, the lesion had gross central necrosis, and dual-phase CT and angiographic findings were consistent with the characteristic features reflecting a necrotic, ruptured hepatic angiosarcoma.

K. Ikeda (✉) · M. Maehara · N. Ohmura · H. Kurokawa · S. Sawada
Department of Radiology, Kansai Medical University, 10-15 Fumizono, Moriguchi, Osaka 570-8507, Japan
Tel. +81-6-6992-1001; Fax +81-6-6991-1249
e-mail: ikedakos@takii.kmu.ac.jp

K. Koda
Department of Pathology, Fujieda Municipal General Hospital, Fujieda, Shizuoka, Japan

H. Yokoyama
Department of Surgery, Fujieda Municipal General Hospital, Fujieda, Shizuoka, Japan

Case report

A 57-year-old man presented with a 1-week history of right back pain. He had no significant past medical history. He did not consume alcohol. On physical examination, the patient was icteric and had a low-grade fever. His abdomen was distended, with a tender palpable liver. Liver function tests were near-normal. Serology was negative for hepatitis B and C. Levels of tumor makers α-fetoprotein and carcinoembryonic atigen (CEA) were within normal limits.

The initial abdominal radiograph showed an upper abdominal mass displacing the bowel loops toward the pelvis, consistent with massive hepatomegaly. Abdominal ultrasonography confirmed the presence of ascites and hepatomegaly with hyperechoic parenchymal foci within the liver. Unenhanced CT demonstrated a right hepatic lobe mass, hypoattenuated to the liver and isoattenuated to the aorta. A high-attenuation intratumoral hemorrhagic liver mass was noted. Arterial

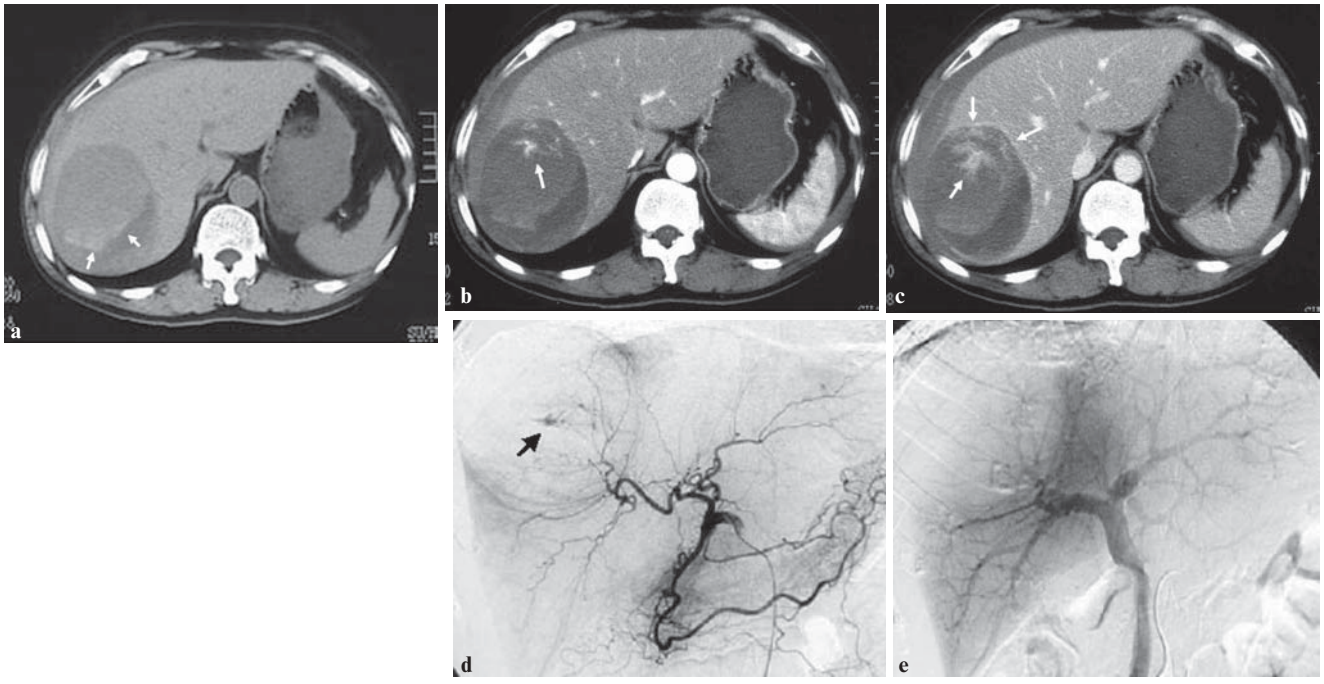

Fig. 1a–e. Solitary angiosarcoma with intratumoral hemorrhage and hemoperitoneum in a 57-year-old man. Note the hemoperitoneum anterior to the right hepatic lobe tumor. **a** Unenhanced computed tomography (CT) scan shows the right hepatic lobe mass, hypoattenuated to the liver and isoattenuated to the aorta. A high-attenuation intratumoral hemorrhagic liver mass (*arrows*) and hemoperitoneum were found. **b** Arterial phase contrast-enhanced CT scan shows the liver mass, hypoattenuated to the liver, with small patchy enhancement isoattenuated to the aorta (*arrow*) and with hypovascular areas reflecting necrosis. **c** Portal venous phase contrast-enhanced CT scan shows that the liver mass was hypoattenuated to the liver and had small areas of patchy enhancement that progressively became isoattenuated to the aorta (*arrows*), reflecting the vascular nature of the tumor. **d** Angiography during the arterial phase reveals a quite small enhanced area of the right hepatic lobe mass (*arrow*); other areas of the hepatic mass are not obviously enhanced, reflecting necrosis in the tumor. **e** Angiography during the portal phase obviously does not reveal an enhanced area of the right hepatic lobe mass

phase contrast-enhanced CT scan showed a liver mass hypoattenuated to the liver with areas of small patchy enhancement isoattenuated to the aorta. A portal venous phase contrast-enhanced CT scan showed that the liver mass remained hypoattenuated to the liver with an extensively enhanced area that over time isoattenuated to the aorta (Fig. 1a).

Abdominal angiography during the arterial phase showed a quite small enhanced area of the right hepatic lobe mass; other areas of the tumor were not obviously enhanced. Angiography during the portal phase did not show an enhanced area of the right hepatic lobe mass (Fig. 1d,e). We explained these findings not by postulating extravasation due to hepatic rapture but by pooling of contrast media in the tumor. During angiography, we embolized 15–20 pieces of Spongel from the right hepatic artery.

At surgery, an 8.6-cm right hepatic lesion was found close to the hepatic capsule. It had apparently ruptured through the capsule and bled into the peritoneum where blood clots were found. On pathology, the tumor was found to be composed entirely of hemorrhagic and necrotic lesions with a fibrous capsule (Fig. 2a). The findings were consistent with hepatic angiosarcoma. Histopathologic examination showed pleomorphic endothelial cells, fibrous septa with atypical endothelial cells, and blood pooling. No Thorotrast deposit was found (Fig. 2b). On immunohistochemical staining, the specimen was positive for CD34 and factor VIII, which originated from the vascular endothelium.

After discharge, the patient developed progressive disease with abdominal discomfort and pedal edema. He died 10 months after his initial presentation. No autopsy was performed.

Discussion

On imaging studies, hepatic angiosarcoma, which is a rare tumor with a peak incidence in older men, is commonly composed of hypervascular lesions.[1] Clinical features are nonspecific, and the prognosis is poor. Ex-

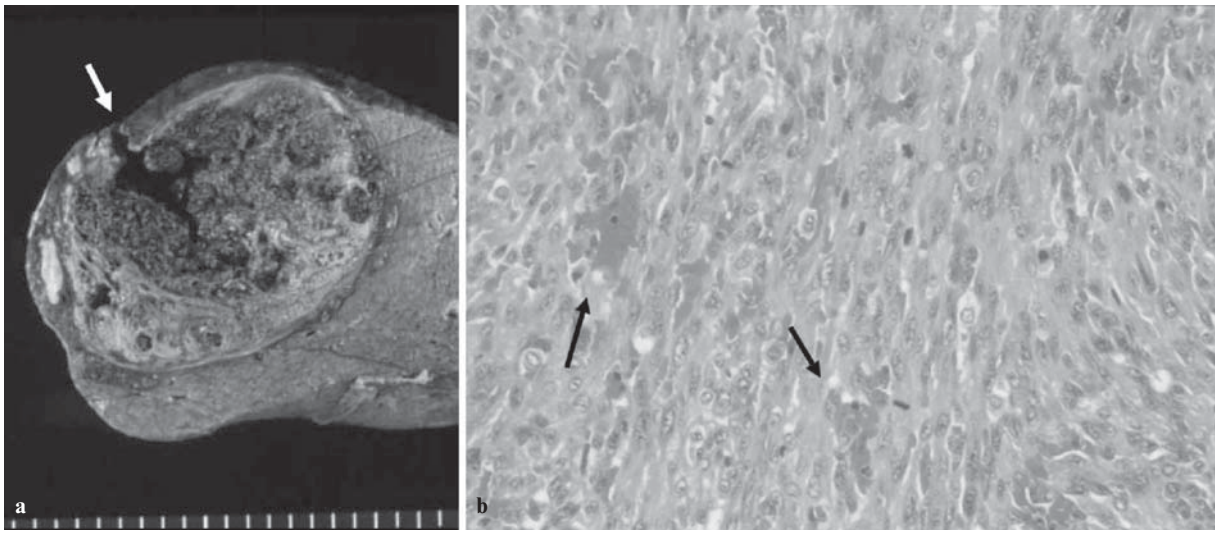

Fig. 2. a Cut section of the gross specimen shows a hemorrhagic mass with a chronic organized subcapsular hematoma. It had apparently ruptured through the capsule (*arrow*). **b** H&E-stained sections show pleomorphic endothelial cells, fibrous septa with atypical endothelial cells, and blood pooling (*arrows*)

posure to a number of carcinogens has been implicated, including thorium dioxide angiographic contrast (Thorotrast), polyvinyl chloride, arsenic, and anabolic steroids. Angiosarcoma has also been associated with hemochromatosis and von Recklinghausen's disease. However, most of the cases are idiopathic.[5] Many patients present with spontaneous hemoperitoneum. Angiosarcoma has reportedly been associated with spontaneous tumor rupture and intraperitoneal hemorrhage.[6] This propensity for hemorrhage probably reflects the vascular nature of the tumor. Tumor rupture and hemoperitoneum are likely caused by tumors arising in subcapsular locations.

On gross inspection, angiosarcomas are spongy, vascular, and hemorrhagic tumors that can range from being well-differentiated lesions with well-formed but irregular anastomosing vessels lined by minimally atypical cells to neoplasms composed of anaplastic spindle or epithelioid cells with few if any vascular lumens. Necrosis and hemorrhage are often present.[7]

The CT appearance of hepatic angiosarcoma has been extensively documented and reflects the variable vascularity of the tumor. Earlier reports that nodular centripetal contrast enhancement on dual-phase CT can mimic cavernous hemangioma highlights a potential diagnostic pitfall.[4,8] However, Peterson et al. reported that only one of six cases in a recent series showed this finding.[9] Hepatic angiosarcoma characteristically shows many areas that are hypodense to the liver during both the arterial and portal venous phases, as well as other areas that are hyperdense during the arterial phase (CT value close to that of the aorta) and isodense or slightly hyperdense during the portal venous phase.[7,9] Our patient's CT showed the former finding, which is consistent with necrosis and hemorrhage, and the latter finding, which is consistent with the vascular nature of the tumor.

The list of vascular primary tumors of the liver is long and includes hemangioma, hepatocellular carcinoma, angiosarcoma, epithelioid hemangioendothelioma, carcinoid, cholangiosarcoma, and hepatoblastoma.[8] However, when a hepatic mass is shown to be a hypovascular lesion, as in our case, a well-differentiated hepatocellular carcinoma (HCC) and a metastases are the two most likely diagnoses.[10] Cirrhosis is the most important finding to narrow the differential diagnosis. More than 80% of HCCs are associated with cirrhotic livers. In a noncirrhotic liver, only 2% of malignancies are primary liver tumors. Most cases have a primary lesion in the lung, pancreas, stomach, breast, or other site.[11] In patients with a suspected angiosarcoma, the most important differential diagnoses include metastases (if multiple lesions are present) and cavernous hemangioma[9] (if a single lesion is present). In rare cases, dual-phase CT and angiography might show the characteristic findings of hemangioma, but angiosarcoma should be suspected if there is rapid enlargement or increasing pain.[9]

Dual-phase CT and angiographic findings can provide sufficient information to allow one to distinguish an angiosarcoma from a hemangioma, as most angiosarcomas have avascular areas that reflect the presence of gross central necrosis.

References

1. Falk H, Herbert J, Crowley S, Ishak KG, Thomas LB, Popper H, et al. Epidemiology of hepatic angiosarcoma in the United States: 1964–1974. Environ Health Perspect 1981;41:107–13.
2. Locker GY, Doroshow JH, Zwelling LA, Chabner BA. The clinical features of hepatic angiosarcoma: a report of four cases and a review of the English literature. Medicine (Baltimore) 1979;58:48–64.
3. Vasile N, Larde D, Zafrani ES, Berard H, Mathieu D. Hepatic angiosarcoma. J Comput Assist Tomogr 1983;7:899–901.
4. Itai Y, Teraoka T. Angiosarcoma of the liver mimicking cavernous hemangioma on dynamic CT. J Comput Assist Tomogr 1989;13:910–2.
5. Rademaker J, Widjaja A, Galanski M. Hepatic hemangiosarcoma: imaging findings and differential diagnosis. Eur Radiol 2000;10:129–33.
6. Mahony B, Jeffrey RB, Federle MP. Spontaneous rupture of hepatic and splenic angiosarcoma demonstrated by CT. AJR Am J Roentgenol 1982;138:965–6.
7. Koyama T, Fletcher JG, Johnson CD, Kuo MS, Notohara K, Burgart LJ. Primary hepatic angiosarcoma: findings at CT and MR imaging. Radiology 2002;222:667–73.
8. White PG, Adams H, Smith PM. The computed tomographic appearances of angiosarcoma of the liver. Clin Radiol 1993; 48:321–5.
9. Peterson MS, Baron RL, Rankin SC. Hepatic angiosarcoma: findings on multiphasic contrast-enhanced helical CT do not mimic hepatic hemangioma. AJR Am J Roentgenol 2000;175: 165–70.
10. Yoshimatsu S, Inoue Y, Ibukuro K, Suzuki S. Hypovascular hepatocellular carcinoma undetected at angiography and CT with iodized oil. Radiology 1989;171:343–7.
11. Melia WM, Wilkinson ML, Portmann BC, Johnson PJ, Williams R. Hepatocellular carcinoma in the non-cirrhotic liver: a comparison with that complicating cirrhosis. Q J Med 1984; 53:391–400.