

A case of giant hepatic hemangioma with spontaneous intratumoral bleeding.

<Case Report>

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ABSTRACT

Cavernous hamangioma is the most common benign tumor of the liver, and most of them remain asymptomatic. However, a giant hemangioma may rupture spontaneously and present severe complications such as shock and anemia, and need emergency operation. We report a case that spontaneous intratumoral bleeding of a giant hemangioma of the liver that occurred in a 55-year-old woman. She presented with severe abdominal pain, anemia, and shock at 3 and a half years after a transcatheter arte-

rial embolization (TAE) treatment. Although the bleeding was under control, a surgical procedure was considered to reduce the risk for re-rupture. The general outcome of surgical treatment for giant hemangiomas is satisfactory. In contrast, especially in our case, TAE was unable to reduce the risk of rupture over the long term. Therefore, surgery for giant hemangioma of the liver should be considered as the first treatment to avoid the risk of rupture even in "healthy" case.

Key words : Cavernous hemangioma, Spontaneous rupture, Intratumoral bleeding

INTRODUCTION

Cavernous hemangioma is the most common benign tumor of the liver,¹⁻³⁾ and the natural history of the hepatic hemangioma remains asymptomatic. In contrast, giant hemangiomas are sometimes symptomatic and severe complications such as jaundice, Kasabach-Merritt syndrome, and spontaneous rupture may occur.⁴⁾ Spontaneous rupture of hemangiomas can result in shock and anemia, and needs emergency op-

eration. Recently, trans-catheter arterial embolization (TAE) has been shown to be effective for the management of rupture of hepatic hemangiomas.⁵⁾ Most cases of ruptured hemangioma rupture in the subcapsule,^{4,6)} and intratumoral bleeding is very rare.⁷⁾ Recently, we encountered a case of giant hepatic hemangioma with spontaneous intratumoral bleeding accompanied by severe pain, shock and anemia.

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CASE REPORT

The patient was a 51-year-old woman suffered from right upper quadrant abdominal pain and vomiting and visited our department for further examination on November 14, 1999. The hematological study showed only slight anemia, and normal AFP, CEA, and CA19-9 levels. US demonstrated a huge heterogeneous tumor in the right lobe of the liver and a small hyperechoic tumor in the left lobe. CT with contrast showed a large tumor with a peripheral nodular pattern of enhancement with a hypodense center in the right lobe (Segment 8) and small tumor in the left lobe (Segment 4) (Fig. 1A). On a hepatic angiogram, stretching of the right anterior and posterior hepatic arteries with a cotton-wool appearance were observed (Fig. 1B). The radiological led to a diagnosis of a hemangioma of the liver 10 cm in a diameter. Transcatheter hepatic arterial embolization (TAE) with a gelatin sponge was performed for the right branch of the hepatic artery (Fig. 1C). Some complications such as abdominal pain and mild fever were noticed after TAE, but no serious complication occurred. After TAE, she was asymptomatic and the size of the hemangioma showed

no change in radiological examination. After 3 and a half years, she suddenly had severe abdominal pain and radiating pain to the right shoulder at midnight. Laboratory studies revealed severe anemia (red blood cells: $289 \times 10^4/\text{mm}^3$; hemoglobin concentration; 7.0 g/dl) and hypovolemic shock. US showed a hyperechoic mass with a spotted echo internally in the right hepatic lobe. CT demonstrated intratumoral hemorrhage of the liver hemangioma (Fig. 2). We considered emergency operation and TAE. However, the anemia had not progressed and she recovered from shock after blood transfusion. After her recovery from anemia and shock, we decided on tumor resection because of the risk of re-rupture. Enucleation of the hemangioma was performed 14 days after the rupture episode. The tumor was on the surface of the right lobe of the liver (Fig. 3A). The resected specimen measured $14.0 \times 13.5\text{cm}$. On a cut section, the tumor consisted of a sponge-like substance and a huge hematoma due to the intracavity bleeding (Fig. 3B). Histopathological examination revealed a cavernous hemangioma composed of dilated vascular spaces of various sizes with thrombus (Fig. 3C).

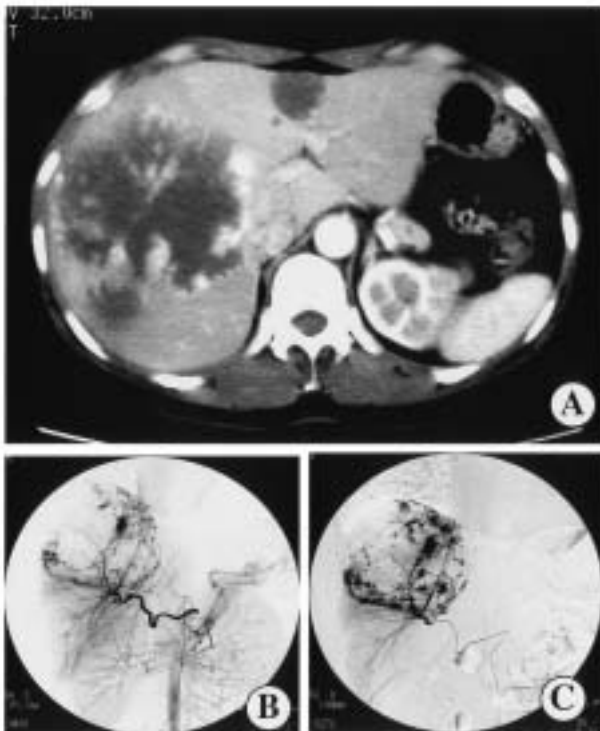
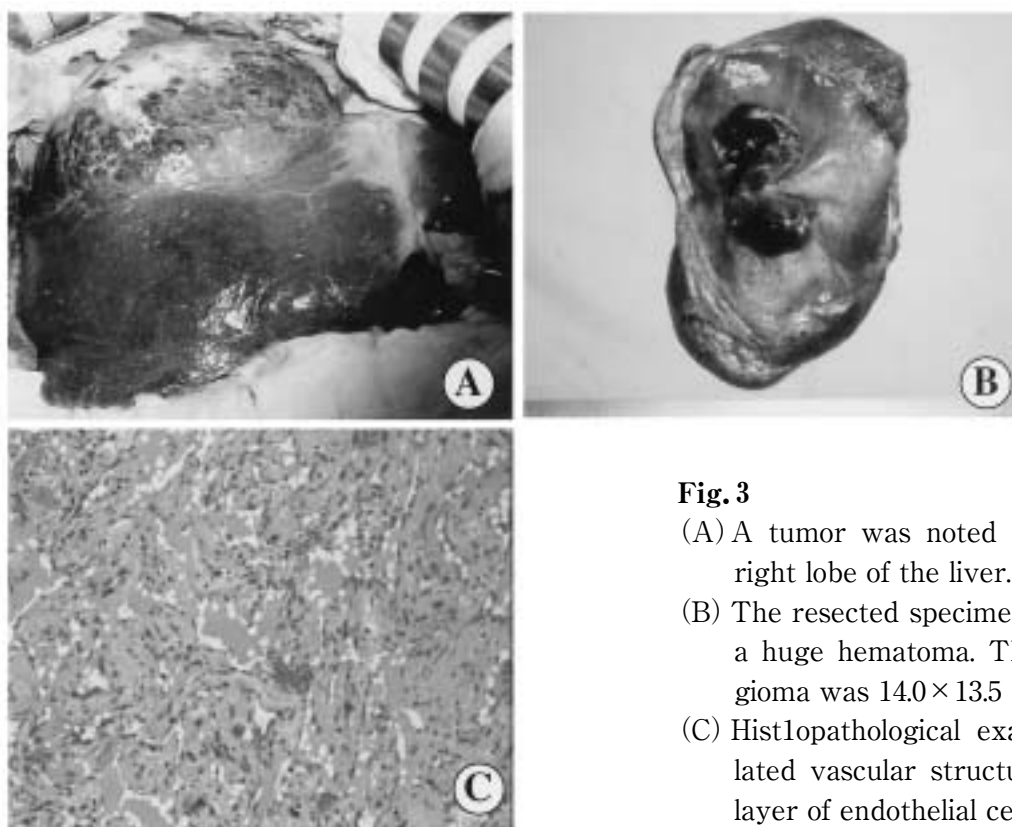


Fig. 1

- (A) Contrast-enhanced CT shows a large tumor in the right lobe of the liver with peripheral enhancement in the nodular areas.
- (B) Hepatic angiogram shows a stretch of the right anterior and posterior hepatic arteries and cotton-wool appearance.
- (C) Transarterial embolization with gelatin sponge was performed in the right branch of the hepatic artery for hemostasis.

**Fig. 2**

Plain CT shows an intratumoral hemorrhage of the liver hemangioma after three and a half years.

**Fig. 3**

- (A) A tumor was noted on the surface of the right lobe of the liver.
 (B) The resected specimen was soft tumor with a huge hematoma. The size of the hemangioma was 14.0 × 13.5 cm.
 (C) Histopathological examination reveals dilated vascular structures lined by a single layer of endothelial cells with thrombus.

DISCUSSION

Cavernous hemangiomas is one of the most common benign tumors of the liver,⁸⁾ and it is usually are small (<4cm), asymptomatic and found incidentally by modern imaging techniques such as US and CT.^{6,9)} The majority of patients are managed conservatively without specific treatment.^{10,11)} However, giant hemangiomas defined as those 4cm in diameter or

greater, may cause pain, abdominal fullness, nausea or vomiting, and surgical resection should be considered.¹²⁾ In some cases, there are severe complications such as jaundice, hemobilia, and Kasabach-Merritt syndrome. Rupture sometimes occurs accompanied by symptoms such as sudden onset of severe abdominal pain, abdominal distention, and hypotension or shock.¹³⁻¹⁶⁾ We have encountered a case of giant

liver hemangioma with intratumoral bleeding. Almost all ruptures of hepatic hemangioma, occur in the subcapsular area, and intratumoral bleeding is very rare.⁷⁾ Our patient had anemia and hypovolemic shock due to intratumoral bleeding. Symptomatic hemangioma is usually large, and needs to be treated by operation or TAE. TAE has been used for treatment of hepatic hemangioma⁶⁾ including intratumoral bleeding, and for patients in poor clinical condition.⁴⁾ Srivastava *et al.* reported the role of TAE as an effective treatment for symptomatic hemangioma that could be used to avoid unnecessary surgery.⁵⁾ Although we considered TAE in our case, the anemia did not progress and the patient recovered from shock after blood transfusion. Thus, we did not need to perform TAE. In this case, spontaneous hemostasis occurred within the limited bleeding volume, because of the site of bleeding was within the tumor. In a sense she was lucky. However, we decided on tumor resection because of the risk of re-rupture. Indications for operation in hemangioma have traditionally been the development of pain, especially in patients with rupture, rapidly enlarging lesions, profound thrombocytopenia, or an uncertain diagnosis for the liver mass.^{17, 18)} A variety of techniques for surgery have been carried out, including anatomical liver resection, enucleation, hepatic artery ligation, and liver transplantation^{19, 20)}. The most common operation methods are liver resection or enucleation. Enucleation is associated with significantly decreased blood loss, preservation of the normal hepatic parenchyma and fewer complications postoperatively compared with anatomic resection.²¹⁾ Although our case showed displacement and compression of the right and middle hepatic vein by the tumor, we could carry out enucleation without damage to the hepatic vein. In resected cases, recurrences of hemangioma are rare,²²⁾ and the outcome of surgery is satisfactory. TAE for giant hemangioma in our case seemed not to reduce the risk of spontaneous hemorrhage in the long term. Therefore, surgery for hepatic giant hemangioma should be

considered to avoid the risk of rupture in "healthy" patients.

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