



A case of spontaneous hepatic hemangioma rupture: Successful management with transarterial chemoembolization alone

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ABSTRACT

Hemangioma is the most common benign hepatic tumor. Although spontaneous rupture is rare, the mortality rate ranges from 60 to 75%. Only 34 cases have been reported in the literature, with only one report using transcatheter arterial embolization (TAE) alone as treatment. We report a case of spontaneous rupture with “flowering sign” of a giant hepatic hemangioma, presenting with acute abdominal pain and shock, while the volume of the hemangioma and blood loss were similar. The patient was successfully managed by transarterial chemoembolization (TACE) alone, which has an operative mortality rate of up to 36.4%.

Introduction

Hepatic hemangioma is the most common benign tumor of the liver. The autopsy occurrence ranges between 0.4 and 7.3%.¹ Although most people with hemangiomas are asymptomatic, there is an uncommon but fatal complication: spontaneous rupture of tumors with or without hemoperitoneum.² There have been 34 cases of spontaneous rupture of hepatic hemangioma reported in the literature³ before the present case, with only one case reported using transcatheter arterial embolization (TAE) alone as treatment, which has shown impressive results at 6-month follow up.³ We report a case of giant hepatic hemangioma accompanied by spontaneous rupture that was well managed with transarterial chemoembolization (TACE) alone and in which we observed an interesting “flowering sign”, with the volume of the hemangioma correlating with that of blood loss.

Case report

A 35-year-old woman experienced sudden onset of diffuse abdominal pain without vomiting and presented to the local hospital immediately. She had no constipation or history of recent trauma. At admission, her hemodynamic parameters were normal. Upon physical examination, she presented upper abdominal sensitivity without any sign of peritoneal irritation. The patient was transferred to the emergency department of our institute the following day. On arrival, patient was pale and in

hemorrhagic shock. Her blood pressure was 81/49 mmHg and her pulse was 58 beats/min and feeble, but still without any sign of peritoneal irritation.

However, a lesion in the right lobe of the liver, with a hybrid echo inside, was detected by ultrasonography, and the color doppler flow imaging showed a blood flow signal around the lesion. Contrast-enhanced computerized tomography (CECT) of the abdomen demonstrated a peripherally enhancing lesion in the right lobe of the liver [Fig. 1 (A-C)].

The results of laboratory tests were as follows: red blood cell count ($2.38 \times 10^{12}/L$), hemoglobin (62 g/L), MCV (84.1 fl), MCH (26.1 pg), MCHC (311 g/L), blood platelet ($457 \times 10^9/L$), white cell count ($8.33 \times 10^9/L$), alanine aminotransferase (44 U/L), aspartate aminotransferase (42 U/L), γ -glutamyl transferase (134 U/L), alkaline phosphatase (240 U/L), and albumin (26 g/L).

The supervising doctor diagnosed the patient with ruptured hepatic hemangioma based on the test results. After initial resuscitation with intravenous fluids and 4 units of packed red blood cells, her vitals were stable before transfer to the interventional operating room for further treatment.

After super-selective catheterization (Terumo Corporation, Japan) was performed on the hemangioma’s feeding artery, digital subtraction angiography (DSA) showed typical features of a ruptured hemangioma with active contrast agent extravasation. The picture of the contrast medium pooling inside the lesion resembled blooming flowers, which we

Abbreviations: TAE, transcatheter arterial embolization; TACE, transarterial chemoembolization; CECT, contrast enhanced computerized tomography.

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Fig. 1. CECT of the abdomen showing a hemangioma in the right lobe of the liver. **(A)** The largest dimension of the lesion was 127 mm in the craniocaudal direction, and the antero-posterior measured 104 mm. **(B)** The blood lake of the hemangioma was clearly seen inside the lesion at the antero-posterior aspect, indicating the presence of active contrast extravasation. **(C)** The transverse section of the CECT of the abdomen measured 126 mm. **(D)** The hemangioma shrunk at the 6-month follow-up, with antero-posterior and transverse measurements of 87 and 93 mm, respectively. CECT: contrast-enhanced computed tomography.

named the “flowering sign” [Fig. 2]. Embolization of the feeding artery of the hemangioma was performed in moderation using 50 mg bleomycin (Terumo Corporation, Japan) lipiodol emulsion and a gelatin sponge (350–500 μm; Cook Bloomington Inc U.S.A) through the same micro-catheter, until the periphery of the hemangioma was completely surrounded.

Seven days later, a postoperative abdominal computerized tomography scan was performed and confirmed that the hemorrhaging had stopped. The laboratory tests on the eighth day indicated that the patient had been returned to the normal ranges. The patient left the hospital on

the ninth day. A 6-month follow-up abdominal computerized tomography scan showed successful regression of the giant hemangioma [Fig. 1(D)], and the patient did not present any discomfort at the 1-year follow-up.

Discussion

The size of a hepatic hemangioma ranges from several millimeters to over 20 cm, with those bigger than 4 cm classified as giant hemangiomas.^{1,4} Symptoms are mostly related to the size and position of the

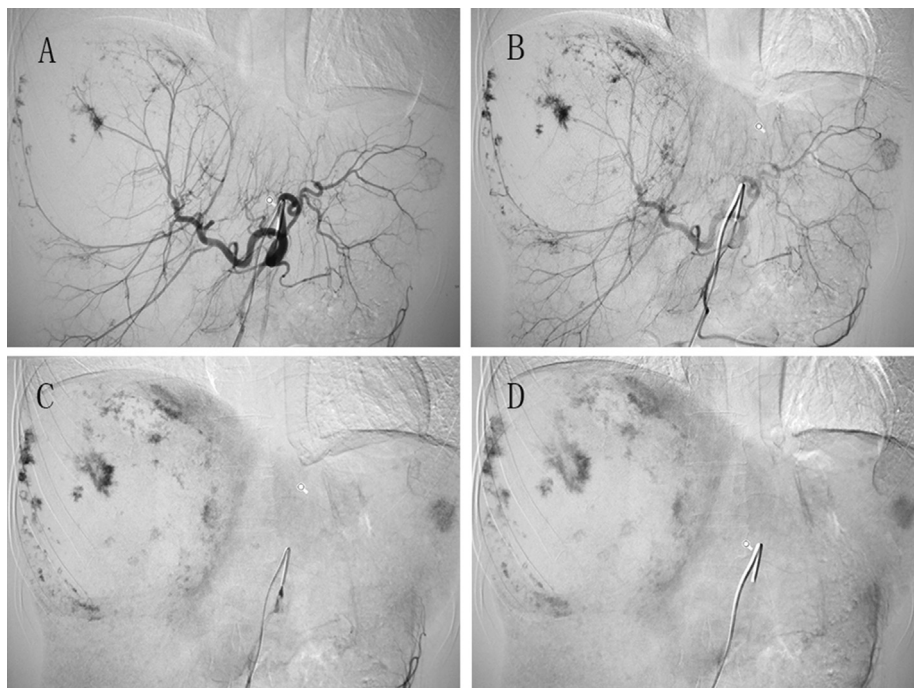


Fig. 2. Digital subtraction angiography showing a hemangioma with active contrast extravasation, similar to blooming flowers, which we named “flowering sign”.

tumor, but most patients are asymptomatic.^{1,7,8} Clinical manifestations of spontaneous rupture of hemangiomas consist of a sudden abdominal pain with or without peritonitis, with one-third of cases having hemodynamic instability and hypovolemic shock due to hemorrhage.³

The diagnosis of hepatic hemangioma can be done through imaging techniques including ultrasonography, dynamic CECT scanning, magnetic resonance imaging, and DSA, which can also be used for treatment.

Spontaneous rupture of hepatic hemangioma is often life-threatening, and the management of complications is challenging as the mortality rate ranges between 60 and 75%,¹ with operative mortality rate accounting for up to 36.4%.⁹ Therefore, recent studies are emphasizing the importance of TAE in the management of hepatic hemangiomas, especially in high risk cases, since the method decreases intraoperative blood loss and significantly improves outcomes.^{3,10} To our knowledge, five cases have been reported that used TAE before surgical resection of a ruptured hepatic hemangioma without operative mortality.¹¹ Furthermore, there were two cases that used TAE/TACE alone to treat spontaneously ruptured hepatic hemangioma,^{3,7} including the present one. In both cases, the patients recovered several days after TAE/TACE and the treated hemangiomas showed significant regression without secondary bleeding or any serious complications during follow-up.⁹

Bleomycin, an antitumor antibiotic, has nonspecific damaging effects on the vascular endothelium. Since the intima of the sinuses of the hemangioma is covered with immature simple endothelium, without muscular or elastic layers, the antitumor agent was used to promote hepatic hemangioma regression in the present case; Zeng et al. previously demonstrated this agent to be a safe and effective treatment.¹² TACE was selected for the patient for three main reasons: firstly, embolization of the hemangioma's feeding artery could prevent bleeding, and sustained bleomycin release from lipiodol would shrink the tumor; secondly, TACE treatment could protect normal liver tissue and minimize surgical trauma since the patient was feeble; finally, reports have demonstrated that TAE/TACE alone is a safe and efficient method for giant hepatic hemangioma.^{3,10} Furthermore, the cost of surgical resection was significant; thus, the management of TACE would be more cost effective for the patient.

There was a "flowering sign" observed in the angiogram. It was postulated that the contrast agent was actively extravasated from the crevasse of the peripheral vessel to the blood pool, due to the blood pressure. This process is similar to the diffusion of a drop of Iodophor instil into water under gravitation, which looks like flowers blooming, and it differs from the characteristic centripetal filling-in of the contrast agent of CECT. As far as we know, we are the first group which has reported this phenomenon.

In addition, there was another interesting phenomenon observed in this case. The volume of the hemangioma was approximately equal to the volume of blood loss, assessed by the loss of hemoglobin in the laboratory test, while there was no other bleeding outside of the tumor. We thus

assumed that the shock of the patient was due to a hemorrhage inside the tumor, which behaved as the "flowering sign"; this hypothesis was similar to that of Saegusa and colleagues.⁶ The rapid expansion of the hemangioma irritated the adjacent liver capsule and lead to the abdominal pain, although this pain occurred without peritonitis; this contributed to the patient recovering in a few days. However, this theory remains to be validated.

In conclusion, we successfully treated a case of spontaneous hepatic hemangioma rupture with TACE alone and demonstrated that TACE alone is a safe and effective method for such cases, even though the operative mortality rate is high.

Conflict of interest

The authors declare no conflict of interest.

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