

# Hemoperitoneum from a Spontaneous Rupture of a Giant Hemangioma of the Liver: Report of a Case

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## Abstract

Hemangioma is the most common benign tumor of the liver and it is often asymptomatic. Spontaneous or traumatic rupture, intratumoral bleeding, consumption coagulopathy, and rapid growth are mandatory surgical indications. We report a case of giant hemangioma of hepatic segments II and III, which presented as hemoperitoneum, and were treated successfully with preoperative transcatheter arterial embolization (TAE) and hepatic bisegmentectomy. A PubMed Medline search has identified up to now 32 cases of spontaneous rupture of hepatic hemangioma in adults (age >14 years) without a history of trauma, including the present case. Twenty-seven out of these were reviewed. Sixteen (84.2%) of 19 tumors of known size were giant hemangiomas (mean diameter 14.8 cm; range 6–25). Twenty-two (95.7%) patients underwent surgery. Thirteen patients (59.1%) had a resection, 5 (22.8%) were sutured, and 4 (18.1%) underwent tamponade. Three (23%) out of the 13 resected patients died. Four patients (30.8%) underwent TAE prior to elective hepatic resection without any operative mortality. Among the 5 sutured patients, 2 (40%) died as well as 3 (75%) out of 4 patients who underwent tamponade. The mortality rate of all surgery patients was 36.4% (8/22).

**Key words** Giant hepatic hemangioma · Hemoperitoneum

## Introduction

Hemangioma is the most common benign tumor of the liver, with a frequency at autopsy of 0.4%–7.3%.<sup>1</sup> In a

large series of 24 000 abdominal ultrasonographies it was diagnosed in 1.7% of cases.<sup>2</sup> The natural history of hepatic hemangioma is not yet well defined, thus leading to several management options. A spontaneous or traumatic rupture, intratumoral bleeding, consumption coagulopathy (Kasabach-Merritt syndrome), and rapid growth are mandatory surgical indications. Persistent abdominal pain, obstructing jaundice, portal hypertension, a size greater than 5 cm, superficial localization because of traumatism risk, and an uncertain diagnosis are relative surgical indications.<sup>2–4</sup> Tumors greater than 4 cm in size are defined as “giant” hemangiomas.<sup>5</sup> At present, the most accepted policy for asymptomatic giant hemangiomas is surveillance.<sup>6,7</sup> Although a rupture is a rare event (1%–4%),<sup>8</sup> it has a high mortality rate (60%–75%),<sup>1</sup> and therefore some surgeons prefer to operate even on asymptomatic giant hemangiomas. The treatment of choice should be based on the balance between the complications that may be prevented by surgical resection and the estimated operative risk of patients.<sup>2</sup>

We report a case of giant hemangioma in the liver, involving segments II and III, which presented as hemoperitoneum due to a spontaneous rupture.

## Case Report

A 53-year-old woman was admitted as a emergency patient to our institution for severe pain in the upper abdomen, which had occurred suddenly without a history of recent traumas. At admission a physical examination revealed severe aching pain in the upper abdomen and signs of peritoneal effusion. Hematochemical tests showed anemia (hemoglobin 8.0 g/dl), slight neutrophilia (7220/mm<sup>3</sup>), and high transaminase level (aspartate aminotransferase 2478; alanine aminotransferase 1205). A computed tomography (CT) scan demonstrated perihepatic and perisplenic effusion, and

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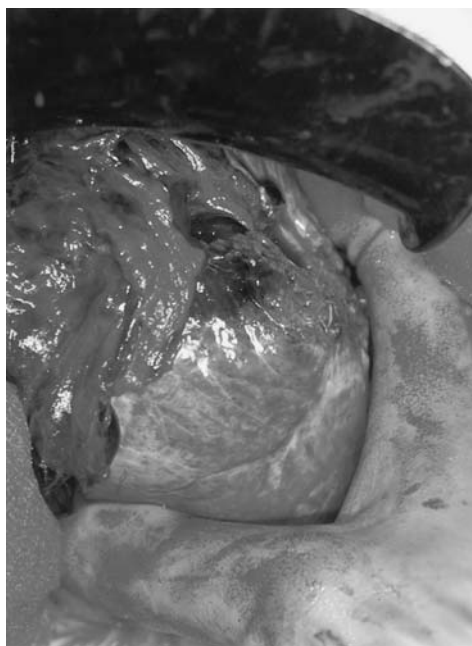


**Fig. 1.** An abdominal computed tomography scan demonstrated a large nonhomogeneous ovoidal mass ( $11.3 \times 8.4$  cm) involving hepatic segments II and III, with contrast enhancement from the periphery to the center, which ruptured on the anterior side and also demonstrated hemoperitoneum

a large ovoidal mass of  $11.3 \times 8.4$  cm, nonhomogeneous, encapsulated, involving the left hepatic lobe, with contrast enhancement from the periphery to the center. An interruption of the capsule was evident anteriorly (Fig. 1). Several other hemangiomas were also present in the remaining hepatic parenchyma. A diagnosis of hepatic angiomatosis with a rupture of the large hemangioma localized in segments II and III was made. Selective hepatic angiography was performed and a large feeding artery was embolized with gelatin sponge particles (Spongostan, Tuttlingen, Germany) (Fig. 2). A temporary standstill of hemorrhage was obtained. On the following day the patient underwent surgery. A right subcostal laparotomy was performed. A large tumor involving segments II and III which was ruptured on the anterior side was found (Fig. 3). In spite of previous embolization, the tumor was actively bleeding into the peritoneum, because of the presence of a feeding accessory left hepatic artery missed at the angiography. After performing the Pringle maneuver and maintaining it for 20 min, a hepatic II–III bisegmentectomy was performed. The histological findings revealed a liver hemangioma of 8.5 cm in diameter. The postoperative course was uneventful and the patient was discharged in the 11th postoperative day. At 1 year follow-up the patient is alive, without any signs or symptoms of recurrence.



**Fig. 2.** Selective left hepatic artery angiography showed a giant hemangioma with a large feeding vessel



**Fig. 3.** An intraoperative picture shows the large tumor involving hepatic segments II and III which was ruptured on the anterior side and was bleeding into the peritoneum

## Discussion

Hemangioma is the most common benign tumor of the liver. The incidence at autopsy ranges between 0.4% and 7.3%.<sup>1</sup> Microscopically, this lesion consists of large vascular spaces lined by a monolayer of endothelial cells, it is well delimited by a surrounding hepatic parenchyma and it has no capsule. Since most hemangiomas are small in size and asymptomatic, they are discovered

during imaging studies for unrelated problems.<sup>2,4,9</sup> Ultrasonography, CT, and magnetic resonance imaging are commonly used for the diagnosis.<sup>2,3,10</sup> A fine-needle aspiration biopsy (FNAB) is not recommended because of the high risk of complications.<sup>4</sup> Laparoscopy in combination with contact ultrasonography may be an effective tool to establish the presence of superficial hemangiomas in patients with an uncertain diagnosis.<sup>2</sup>

According to Adam et al., hemangiomas are defined as “giant” if their diameter exceeds 4 cm.<sup>5</sup> Absolute surgical indications for the hepatic hemangioma are rupture with hemoperitoneum, as in our case, intratumoral bleeding, rapid growth, and consumption coagulopathy (Kasabach-Merritt syndrome). Furthermore, persistent abdominal pain, obstructive jaundice, or portal hypertension, tumors greater than 5 cm or superficially located with a risk of traumatism, and uncertain diagnosis represent relative surgical indications.<sup>2-4</sup> A PubMed Medline (National Library of Medicine) search identified 32 cases of spontaneous rupture of hepatic hemangioma in adults (age >14 years) without a history of trauma, including the present case.<sup>4,8,11-23</sup> Five cases

have been left out from our review because the data were not available. The remaining 27 cases have been reviewed (Table 1) and consist of 17 females and 9 males (in 1 case the gender is not specified), with an average age of 48.2 years (range 15–73; in 2 cases age was not specified). The lesion was located in the right lobe in 14 cases (51.8%), in the left lobe in 10 cases (37%), in both lobes in 1 case (3.7%), and unknown in two. The tumor size ranged from 3 cm to the whole right hepatic lobe. Sixteen (84.2%) out of 19 tumors of known size were giant hemangiomas with a mean diameter of 14.8 cm (range 6–25 cm). Four (14.8%) out of 27 patients died of hemorrhagic shock without any surgery (the outcome of 1 patient was not reported). Of the remaining 23 patients, 22 (95.7%) underwent surgery (in 1 case the management was not specified). Thirteen patients (59.1%) were resected, 5 patients (22.8%) were sutured, and 4 (18.1%) underwent tamponade. Of the 13 patients who underwent a resection, 3 (23%) died. Four patients (30.8%), including our patient, underwent transcatheter arterial embolization (TAE) prior to an elective hepatic resection without any operative

**Table 1.** Twenty-seven cases of a spontaneous rupture of a hepatic giant hemangioma in adults reviewed from a PubMed Medline search

No.	First author <sup>Ref.</sup>	Year	Age (years)	Sex	Tumor size	Tumor location	Management	Outcome
1	Van Haefen <sup>24</sup>	1898	70	F	n.r.	R	<sup>a</sup>	Died
2	Turner <sup>25</sup>	1922	n.r.	M	n.r.	n.r.	Resection	Died
3	Karp <sup>26</sup>	1931	39	F	n.r.	L	Tamponade	Died
4	Tinker <sup>27</sup>	1935	65	F	n.r.	L	Resection	Alive
5	Marckstadt <sup>28</sup>	1938	27	F	Orange-size	L	Tamponade	Died
6	Dahle <sup>29</sup>	1939	44	F	Orange-size	R	Tamponade	Alive
7	Caldwell <sup>30</sup>	1950	61	M	n.r.	L	Suture	Alive
8	Oribe <sup>31</sup>	1951	47	M	Egg-size	R	Tamponade	Died
9	Kruppa <sup>32</sup>	1951	15	F	14 cm	R	Suture	Alive
10	Swell <sup>33</sup>	1961	21	F	10 cm	R	Suture	Died
11	Dessoff <sup>34</sup>	1967	53	M	Whole of right lobe	R	<sup>a</sup>	Died
12	Adam <sup>5</sup>	1970	46	F	25 cm	R	Resection	Died
13	Arnesjo <sup>14</sup>	1975	55	F	10 cm	R	Resection	Died
14	Ong <sup>15</sup>	1975	43	F	Whole of right lobe + IV segment	R-L	Resection	Alive
15	Nyman <sup>35</sup>	1978	37	M	n.r.	L	Suture	Alive
16	Coca <sup>20</sup>	1979	n.r.	n.r.	n.r.	L	Resection	Alive
17	Shiebold <sup>36</sup>	1980	43	F	20 cm	R	Suture	Died
18	Starzl <sup>37</sup>	1980	61	F	15 cm	R	Resection	Alive
19	Dib <sup>17</sup>	1984	44	F	3 cm	n.r.	<sup>a</sup>	Died
20	Nanbu <sup>38</sup>	1986	46	M	4 cm	R	<sup>a</sup>	Died
21	Yamamoto <sup>11</sup>	1991	71	M	6 cm	L	TAE + Resection	Alive
22	Mazziotti <sup>16</sup>	1995	73	M	Giant	R	TAE + Resection	Alive
23	Soyer <sup>18</sup>	1995	36	F	n.r.	R	TAE + Resection	Alive
24	Moreno <sup>4</sup>	1996	68	F	15 cm	L	Resection	Alive
25	Scribano <sup>12</sup>	1996	44	F	Giant	L	n.r.	n.r.
26	Cappellani <sup>8</sup>	2000	43	M	22 cm	R	Resection	Alive
27	Corigliano	2002	53	F	11 cm	L	TAE + Resection	Alive

n.r., not reported; L, left hepatic lobe; R, right hepatic lobe; TAE, transcatheter arterial embolization

<sup>a</sup> Died of hemorrhagic shock without any surgery

mortality. Among the 5 sutured patients, 2 (40%) died as well as 3 (75%) out of the 4 patients who underwent tamponade. The mortality rate for all surgery patients was 36.4% (8/22). The natural history of this tumor is still not well understood and its evolution is unpredictable. No clear therapeutic indications have been established for asymptomatic lesions. Yamagata et al.,<sup>39</sup> in a series of 20 hepatic hemangiomas, 95% greater than 5 cm in diameter, did not demonstrate a size increase, rupture, or progression of symptoms at a mean follow-up of 47 months (range 18–96 months). At a mean follow-up of 5.9 years (range 1.4–12.6 years), Moreno et al.<sup>4</sup> reported in 1 patient (9%), out of 11 patients affected by hepatic hemangioma, a growth from 7.4 to 9.6 cm and occurrence of symptoms. Using ultrasonography, Pietrabissa et al.<sup>2</sup> studied, for a mean follow-up of 55 months (16–72 months), 36 cases of hepatic hemangioma, and observed an unmodified size in 32 cases (88.9%), a slight increase in size (0.5 cm) in 3 cases (8.3%), and a decrease in the diameter of 0.6 cm in 1 case (2.8%). He concluded that lesions exceeding 10 cm in diameter may have greater internal bleeding and further growth or rupture, thus justifying a preventive excision though they were asymptomatic. However, the factors influencing growth or causing a spontaneous rupture are still unknown.

When surgery is indicated, enucleation with temporary inflow occlusion (Pringle maneuver) is the treatment of choice, and it is associated with fewer postoperative complications and less blood loss when compared with anatomic resection.<sup>3</sup> Furthermore, it helps to reduce the loss of the normal liver parenchyma.<sup>2</sup> The presence of a pseudocapsule created by the compression of the surrounding parenchyma permits this surgical technique. The tumor is easily cleaved from the plane of the pseudocapsule, because it is crossed by few vessels and biliary ducts.<sup>10</sup> Occasionally, the tumor location or the involvement of a whole lobe precludes a safe enucleation, and at such times an anatomic resection should be performed.<sup>2,3</sup>

In cases of a rupture, as in our case, TAE is considered to be a valid procedure in stanching or reducing the hemorrhage, thus making it a safer hepatic resection.<sup>5</sup> Suzuki et al.<sup>9</sup> suggested the use of preoperative TAE in patients with consumption coagulopathy related to intravascular coagulation in the hemangioma. He observed an improvement in coagulative factors and a decrease in the intraoperative blood loss, when compared with patients treated only with preoperative intravenous administration of heparin and gabexate mesylate.

In conclusion, surgery is mandatory for ruptured or bleeding hepatic hemangiomas. Furthermore, surgical treatment should also be considered for rapidly growing lesions and symptomatic patients. At present, a poor

knowledge of the natural history justifies the observation management for all asymptomatic hemangiomas. In cases which are diagnostically unclear, laparoscopy in combination with laparoscopic contact ultrasonography may be a useful tool to confirm a diagnosis of hemangioma.

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