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Spontaneous Rupture of Giant Liver Hemangioma: Case Report

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SUMMARY

Introduction Hemangioma is the most frequent benign solid tumor of the liver. It is well known that a giant liver hemangioma carries the risk of spontaneous rupture, followed by hemoperitoneum and hemorrhagic shock with possible fatal outcome.

Case Outline This is a case report of the spontaneous rupture of a giant cavernous hemangioma of the liver in an 85-year old patient. The patient was presented with abdominal pain and hemorrhagic shock. Emergency ultrasonography and computed tomography of the abdomen showed a heterogeneous ruptured solid tumor of the right liver lobe, multiple cysts in the left lobe and massive hemoperitoneum. The patient was successfully managed by immediate exploratory laparotomy, surgical enucleation of the hemangioma under intermittent inflow vascular occlusion, temporary perihepatic packing and planned second look relaparotomy.

Conclusion Immediate surgical procedure is indicated mandatory in unstable patients with a ruptured giant hemangioma of the liver. Surgical enucleation under intermittent inflow vascular occlusion and temporary perihepatic packing could be a life-saving procedure in those patients.

Keywords: liver hemangioma; spontaneous rupture; hemoperitoneum

INTRODUCTION

Hemangioma is the most frequent benign solid tumor of the liver with 52% of all, often present in younger people, predominantly in the fifth decade, with overall prevalence in the general population estimated to range between 0.4-20% [1-5]. There are also reports that women are predominantly affected (4:1 to 6:1) [6]. Hemangioma may be defined as benign vascular tumors with unknown etiology, originates from the primary mesenchymal cells [4]. Histologically, hemangioma can be capillary or cavernous and lesion is consisted of hyperplastic vascular channels covered inside by the vascular endothelium [7, 8, 9].

Adam and coworkers classified hemangiomas as giant if their diameter exceeded 4 cm [2]. Even giant hemangiomas frequently remain asymptomatic; rare but most severe, even fatal complication of the giant hemangioma, is spontaneous rupture (1–4%) [2, 7, 10, 11]. This disastrous event is due to a sudden life-threatening hemorrhage in the peritoneum, with mortality ranging from 60–75% [3, 12-15].

The first case of rupture of the liver hemangioma was reported by Van Haefen [16] in 1898, in a 70-year old woman who died of hemorrhagic shock. In the case of ruptured hemangiomas, followed by serious intraperitoneal hemorrhage, the number of diagnostic procedures is perpetually diminishing, reducing them to immediate ultrasonography, emergency computed tomography (CT scan), peritoneal lavage and laparoscopy [17]. A spontaneous rupture of the giant hemangioma would have possible disastrous outcome if not promptly managed; hemostasis is the first therapeutical option [18].

CASE REPORT

This article reports a case of spontaneous rupture of a giant cavernous hemangioma of the liver in an 85-year old patient who presented with abdominal pain and hemorrhagic shock. He was admitted as an emergency patient to the Clinic for Emergency Surgery of the Clinical Center of Serbia. The patient denied previous abdominal trauma. He suffered from unstable arterial hypertension and cardiomyopathy; and had undergone three earlier abdominal operations due to adhesive intestinal obstruction and incarcerated groin hernia.

On admission, the patient showed hemodynamic instability (pulse >112 times/minute, systolic pressure <84 mmHg), tenderness and muscular rigidity over the upper abdomen. Hemoglobin value of 84 g/L, hematocrit of 0.25, leukocytosis (WBC=14×10^9/l), with elevated serum transaminase values: aspartate aminotransferase (AST=1696 U/L) and alanine aminotransferase (ALT=1587 U/L). The abdominal ultrasound recorded a significant

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amount of free fluid and a giant tumor in the right lobe of the liver. Emergency CT scan confirmed findings of hemoperitoneum, measured reduction of 77 HU (Hounsefield Units) suggested fluid blood as well as blood clots. A large nonhomogeneous mass in the liver was present occupying segments 6, 7, 8. The dimension of the lesion was 11.3 cm in the craniocaudal direction and transversal measure was 8.4 cm (Figure 1). The remaining liver parenchyma was full of multiple simple cysts up to 5 cm in diameter.

After emergency laparotomy, a large amount of blood (1800 ml) was evacuated from the peritoneal cavity. Exploration showed a ruptured and massively bleeding huge tumor in the right lobe of the liver (6, 7, 8), while the remaining liver was full of simple cysts. The patient had spontaneous rupture of the giant cavernous hemangioma of the liver. An atraumatic vascular clamp was placed across the hepatoduodenal ligament and intermittent Pringle maneuver was used in cycles of 15/5 min of clamp/unclamp times.

Under vascular control we performed enucleation of the hemangioma using the ultrasonic dissector (CUSA) with sutured blood vessels and bile ducts. The patient received 8 units (300 ml each) of the whole blood and 15 units of fresh frozen plasma. Control of hemostasis showed no more bleeding. Finally, a careful but sufficient perihepatic five packages of gauze tamponade was placed between the liver, right diaphragm and abdominal wall (perihepatic packing). Abdominal drainage tubes were inserted into the abdominal cavity and the laparotomy incision was sutured.

Macroscopically, the fragments of the tumor were soft, red, and often had firm gray-white streaks or areas. A definitive pathologic examination of the specimen revealed a cavernous hemangioma. Organizing thrombi were common, and focally obliterated the vessels in parts of the tumor. In the neighboring liver tissue, a mild lymphocytic infiltration of sinusoids was detected adjacent to the hemangioma.

On postoperative day 3 (after 72 hours), a planned second look laparotomy was performed. The gauze perihepatic package was removed; no more active bleeding or bile leakage was seen and the laparotomy was definitely closed. During early postoperative course in the Intensive Care Unit (ICU), the principles of aggressive fluid circulatory volume resuscitation guided by monitoring of the central venous pressure and urinary output were applied. On the first postoperative CT scan 7 days after operation, there was a visible hematoma at the place of previous lesion. There were no others hemangiomas, while the remaining hepatic parenchyma was full of simple cysts (Figure 2). Hemoglobin value was 110 g/l, hematocrit 0.34 and serum transaminase level was AST=36 and ALT=50.

The patient was successfully recovered and he was discharged on postoperative day 11.

Follow-up CT scan, performed three months later, showed a complete restitution and moderate hypertrophy of the right lobe with a small scar on the site of previous ruptured hemangioma (Figure 3).

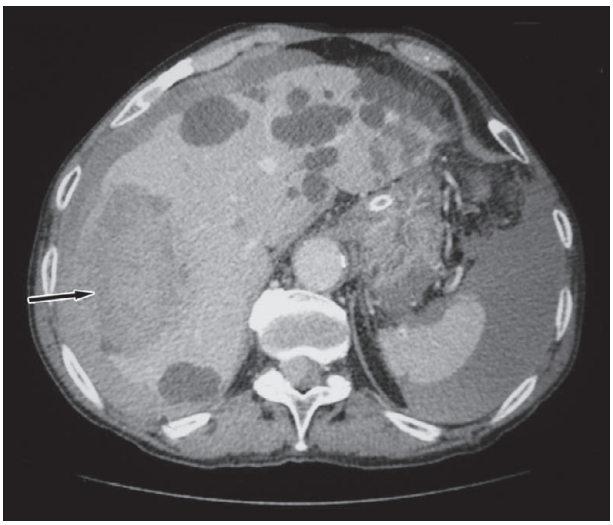

Figure 1. Emergency CT scan performed on admittance. A huge, clearly visible ruptured hemangioma positioned in the right lobe of the liver (arrow). Also visible leakage and blood accumulation surrounding the liver indicating hemoperitoneum. Multiple cysts present in the remaining hepatic parenchyma.

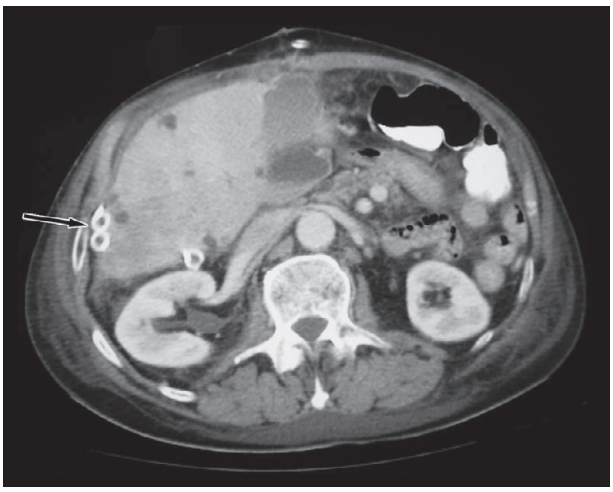

Figure 2. CT scan performed 7 days after operation. Visible a lesser hematoma in the site of previous hemangioma, as well as two abdominal drains, still placed in the perihepatic space (arrow).

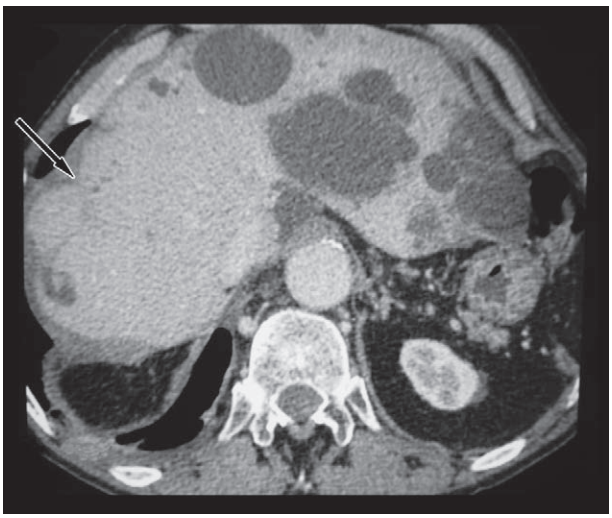

Figure 3. A control postoperative CT scan performed three months later shows moderate hypertrophy of the right lobe of the liver. Also visible a small scar at the site of previous hemangioma, with no signs of recurrence (arrow). Multiple cysts presented in the remaining liver.

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DISCUSSION

Hemangioma is the most frequent benign liver tumor [2, 19]. There are well-circumscribed hypervascular lesions with a clear sheath of compressed liver parenchyma between the hemangiomatous tissue and normal liver. Spontaneous rupture of a giant liver hemangioma is not a usual complication but can be a dramatic event [7, 11]. Without prompt and adequate treatment in that situation patients can die of massive hemorrhage [7, 11].

The first case of rupture of liver hemangioma was reported by Van Haefen [16] in 1898. Reviewing the available literature data, a total of 30 cases of spontaneous ruptures of liver hemangioma have been reported [9] (Table 1). Pietrabissa et al. [17] followed 78 patients during the median period of 55 months (16-72), and concluded that only hemangiomas larger than 10 cm carry a great risk of rupture. Moreno et al. [20] followed 11 patients with liver hemangioma during average period of 6 years (1.4-12.6 years). The growing of tumor, but without rupture was recorded only in one case. Yamagata et al. [21] followed 20 patients with liver hemangioma between 18-96 months (median period 47 months) and he found no progression, growing or rupture of the tumor.

During the last few decades, the imaging methods of hemangiomas have been significantly improved. Radionuclide scanning, ultrasonography, computerized spiral multi-slice tomography (CT), nuclear magnetic resonance (MRI), selective angiography and single photon emission computed tomography (SPECT) can be helpful [22, 23].

The best treatment for a ruptured giant hemangioma of the liver is still controversial [7]. A major complex of the interventional radiological techniques, such as transcatheter arterial embolization, embolization of portal vein, or transhepatic venous stenting, have become more widely used and are now an integral part of the management of these lesion [7, 10, 12]. On the other hand, surgical procedures such as enucleation, segmentectomy, sectorectomy, lobectomy or liver transplantation are also in use [1, 4, 5, 6, 11, 12, 14, 24-28].

Surgical management of the giant ruptured hemangioma followed by hemorrhagic shock in unstable patients is always a great challenge [24-28]. The decision for the method of choice must be gently and carefully balanced between sufficient operative hemostasis and possible postoperative complications [9]. Whatever is the surgeon's decision, the larger the size of the hemangioma, the more difficult the achievement of hemostasis [5]. Pringle maneuver

Table 1. Review of thirty cases with spontaneous ruptures of giant liver hemangioma, modified according to Corigliano [9]

No.	Author [reference no.]	Year	Age (years)	Sex	Size of hemangioma	Localization	Management	Outcome
1	Van Haefen [16]	1898	70	F	Unknown	Right lobe	0	Died
2	Turner	1922	Unknown	M	Unknown	Unknown	Resection	Died
3	Karp	1931	39	F	Unknown	Left lobe	Tamponade	Died
4	Tinker	1935	65	F	Unknown	Left lobe	Resection	Alive
5	Marckstadt	1938	27	F	Orange size	Left lobe	Tamponade	Died
6	Dahle	1939	44	F	Orange size	Right lobe	Tamponade	Alive
7	Caldwell	1950	61	M	Unknown	Left lobe	Suture	Alive
8	Oribe	1951	47	M	Egg size	Right lobe	Tamponade	Died
9	Kruppa	1951	15	F	14 cm	Right lobe	Suture	Alive
10	Swell	1961	21	F	10 cm	Right lobe	Suture	Died
11	Dessoff	1967	53	M	Whole right lobe	Right lobe	0	Died
12	Adam [2]	1970	46	F	25 cm	Right lobe	Resection	Died
13	Arnesjo	1975	55	F	10 cm	Right lobe	Resection	Died
14	Ong	1975	43	F	Giant	Right lobe + seg. 4	Resection	Alive
15	Nyman	1978	37	M	Unknown	Left lobe	Suture	Alive
16	Coca	1979	Unknown	Unknown	Unknown	Left lobe	Resection	Alive
17	Shiebold	1980	43	F	20 cm	Right lobe	Suture	Died
18	Starzl [19]	1980	61	F	15 cm	Right lobe	Resection	Alive
19	Dib	1984	44	F	3 cm	Unknown	0	Died
20	Nanbu	1986	46	M	4 cm	Right lobe	0	Died
21	Yamamoto [10]	1991	71	M	6 cm	Left lobe	TAE + resection	Alive
22	Mazziotti	1995	73	M	Giant	Right lobe	TAE + resection	Alive
23	Soyer	1995	36	F	Unknown	Right lobe	TAE + resection	Alive
24	Moreno [20]	1996	68	F	15 cm	Left lobe	Resection	Alive
25	Scribano	1996	44	F	Giant	Left lobe	Unknown	Unknown
26	Cappellani [7]	2000	43	M	22 cm	Right lobe	Resection	Alive
27	Corigliano [9]	2002	53	F	11 cm	Left lobe	TAE + resection	Alive
28	Griffa [27]	2005	84	F	Giant	Left lobe	Resection	Alive
29	Santos Rodrigues [28]	2010	25	F	Giant	Right lobe	Resection	Alive
30	Jain [12]	2010	31	M	Giant	Right lobe	TAE + resection	Alive

F – female; M – male; 0 – died of hemorrhagic shock without any surgery; TAE – transcatheter arterial embolization

and perihepatic packing may be recommended to allow control of bleeding and hemodynamic stabilization of the patient [9].

Considering the benign nature of the disease, presently there has been a general consent that enucleation is better than anatomical resection in terms of sparing the liver parenchyma and decreasing the intraoperative blood loss [5, 9, 17, 24]. In case of hemodynamically unstable patients with spontaneous ruptures of liver hemangioma enucleation is the preferred operation and can be performed rapidly and safely, with a potentially lower operative morbidity.

This is the case of the spontaneous rupture of the liver hemangioma followed by hemoperitoneum with hemorrhagic shock successfully treated by emergency laparotomy. We performed enucleation of the hemangioma under intermittent inflow pedicle occlusion (Pringle maneuver), temporary perihepatic packing and second look relaparotomy.

Some recent studies have reported a significant decrease of operative mortality from ruptured giant liver hemangiomas up to 36.4% [9]. Other authors report that mortality in surgical management of giant liver hemangiomas in high volume hepatobiliary-transplant centers, even if they are 40 cm in size, is unusual [4]. Opposite to the latter, it is noteworthy that a significant risk of intraoperative bleeding and postoperative complications may follow any hepatic resection, even in the experienced hands [25, 26].

In conclusion, spontaneous rupture of giant hemangiomas of the liver, followed by hemoperitoneum with hemorrhagic shock would have catastrophic outcome if they were not promptly managed upon surgery. In patients with poor general condition and hemodynamic instability, the treatment of choice may be emergency laparotomy, enucleation of the hemangioma under intermittent inflow vascular occlusion (Pringle maneuver), temporary perihepatic packing and second look relaparotomy.

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Спонтана руптура џиновског хемангиома јетре – приказ болесника

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КРАТАК САДРЖАЈ

Увод Хемангиом је најчешћи бенигни тумор јетре. Познато је да су џиновски хемангиоми јетре праћени ризиком од спонтане руптуре с развојем хематоперитонеума, хеморагичним шоком и могућим смртним исходом.

Приказ болесника Приказан је случај спонтане руптуре џиновског хемангиома јетре код болесника старог 85 година. Он се жалио на бол у трбуху, а развио се и хеморагични шок. Хитан преглед трбуха ултразвуком и компјутеризованом томографијом показали су руптуру хетерогеног тумора у десном лобусу јетре, вишеструке цисте у левом лобусу јетре и масивни хематоперитонеум. Болесник је успешно оперисан: урађене су хитна лапаротомија, енуклеација хемангиома под интермитентном васкуларном оклузијом, привремена перихепатична тампонада и планирана релапаротомија.

Закључак Код болесника нестабилног стања с руптуром џиновског хемангиома јетре индикована је хитна хируршка интервенција. Енуклеација хемангиома под интермитентном васкуларном оклузијом и перихепатична тампонада могу бити процедуре које спасавају живот овим болесницима.

Кључне речи: хемангиом јетре; спонтана руптура; хематоперитонеум

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