—Case Reports—

A Symptomatic Giant Hepatic Hemangioma Treated with Hepatectomy

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Abstract

We describe a patient with symptomatic giant hepatic hemangioma treated with hepatectomy. A 53-year-old woman presented with upper abdominal distension and appetite loss. The medical history included multiple hepatic hemangiomas that had been detected 2 years earlier but were left untreated. Initial laboratory tests revealed pancytopenia and mild coagulopathy. Computed tomography and magnetic resonance imaging demonstrated a giant hemangioma, 27 cm in diameter, in the enlarged right lobe of the liver. The inferior vena cava was compressed by tumor without thrombus in the infrahepatic vena cava. The portal venous phase of supramesenteric arteriography revealed compression of the portal vein. There were several hemangiomas in the left lobe. Gastric outlet obstruction due to giant hepatic hemangioma in the right lobe was diagnosed. Laparotomy was performed, and a markedly enlarged liver was detected. Right hepatectomy was performed with an anterior approach. The liver-hanging maneuver could not be performed because of tumor compression of the inferior vena cava. Right hepatectomy was performed with intermittent clamping (Pringle maneuver). Hepatic hemangiomas of the left lobe were not resected because the remnant liver would be reduced. The weight of the resected specimen was 2,100 g. Pathologic examination of the surgical specimen confirmed the presence of benign hepatic hemangiomas. The postoperative course was uneventful, and the patient's appetite improved. The patient was discharged 8 days after the operation. Abdominal distension decreased and laboratory data improved after the operation. Computed tomography revealed hypertrophy of the left lobe of the liver after the operation.
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Key words: hemangioma, hepatectomy, giant, symptomatic

Introduction

Hepatic hemangiomas are the most frequent hepatic tumors, usually found incidentally during abdominal imaging procedures. Most of the lesions are asymptomatic, but occasionally various complications may be present, especially in patients with large lesions. Surgical therapy has recently become popular, and some recent publications have shown excellent results after operative treatment[1,2].

We describe a patient with a symptomatic giant

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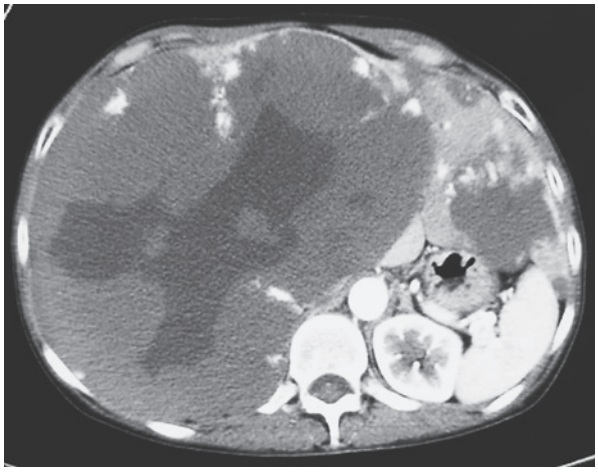

Fig. 1 Computed tomography demonstrated a giant hemangioma, 27 cm in diameter, in the enlarged right lobe of the liver. The inferior vena cava was compressed by a tumor without thrombus in the infrahepatic vena cava.

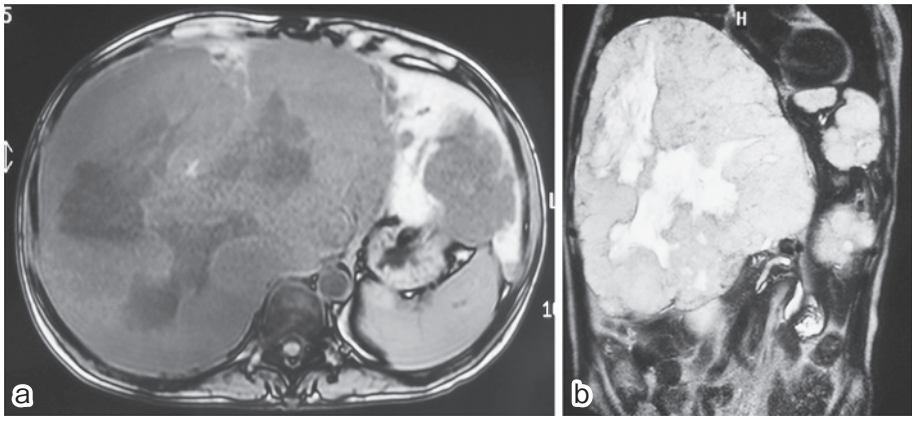

Fig. 2 Magnetic resonance imaging revealed a giant hemangioma, 27 cm in diameter.

hepatic hemangioma treated with hepatectomy.

Case Report

A 53-year-old woman presented with upper abdominal distension and appetite loss. The medical history included multiple hepatic hemangiomas that had been detected 2 years earlier but were left untreated. Initial laboratory tests revealed the following: serum aspartate aminotransferase, 27 IU/L (normal, <28 IU/L); serum alanine aminotransferase, 17 IU/L (normal, <33 IU/L); serum alkaline phosphatase, 185 IU/L (normal 66 to 220 IU/L); serum lactic dehydrogenase, 134 IU/L (normal, 180 to 460 IU/L); serum gamma glutamic transpeptidase, 96 IU/L (normal, 8 to 39 IU/L); total serum bilirubin 1.2 mg/dL (normal, 0.2 to 1.2 mg/dL); serum C-reactive protein, 0.07 mg/dL (normal, <0.3 mg/dL); white blood cell count, 2,600 /μL (normal, 4,000 to 8,000 /μL); red blood cell count, 354 × 10^4 /μL (normal, 410 to 550 × 10^4 /μL); serum hemoglobin concentration, 11.6 g/dL (normal, 14 to 18 g/dL); serum platelet count, 11.3 × 10^4 /μL (normal, 15 to 35 × 10^4 /μL); serum fibrinogen degradation products, 14.3 μg/mL (normal, <10 μg/mL); prothrombin time, 96.7% (normal, 80% to 100%); serum fibrinogen, 141 mg/dL (normal, 200 to 400 mg/dL); D-dimer, 6.04 μg/mL (normal, 0.1 to 1 μg/mL); and antithrombin III, 104.6% (normal, >82%). The serum concentration of carcinoembryonic antigen was 4.6 ng/mL (normal, <2.5 ng/mL), that of CA19-9 was 9.8 u/mL (normal, <37 u/mL), and that of alpha-fetoprotein was 5.1 ng/

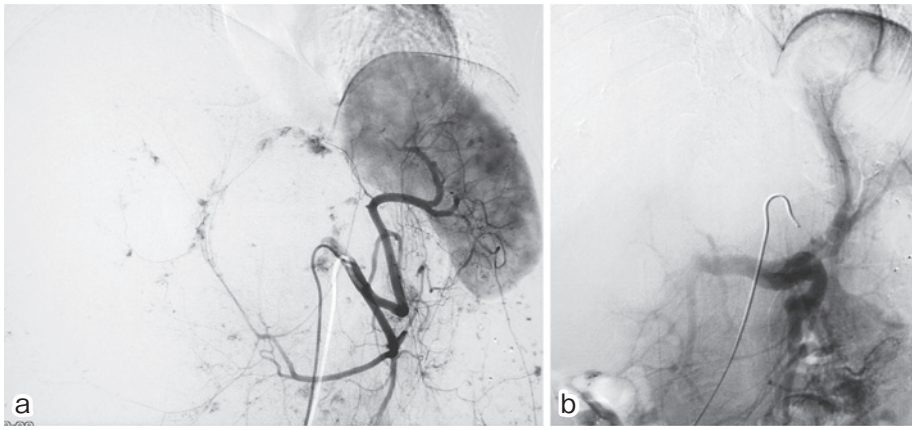

Fig. 3 Celiac arteriography revealed a hypervascular tumor with pooling of contrast medium in the delayed phase. The portal venous phase of supramesenteric arteriography revealed compression of the portal vein.

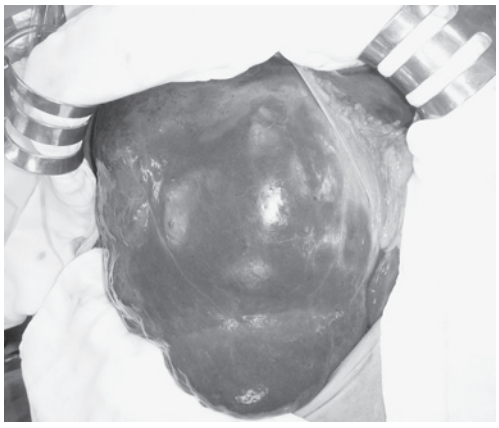

Fig. 4 A markedly enlarged liver was detected.

mL (normal, <10 ng/mL). Computed tomography and magnetic resonance imaging demonstrated a giant hemangioma, 27 cm in diameter, in the enlarged right lobe of the liver. The inferior vena cava was compressed by tumor without thrombus in the infrahepatic vena cava (**Fig. 1, 2**). Celiac arteriography revealed a hypervascular tumor with pooling of contrast medium in the delayed phase. The portal venous phase of supramesenteric arteriography revealed compression of the portal vein (**Fig. 3**). There were several hemangiomas in the left lobe. Gastric outlet obstruction due to giant hepatic hemangioma in the right lobe was diagnosed.

Laparotomy was performed, and a markedly enlarged liver was detected (**Fig. 4**). We tried to mobilize the right lobe of the liver from the diaphragm, but doing so was difficult because of the giant tumor. Therefore, we selected the anterior approach for right hepatectomy. The liver-hanging maneuver could not be performed because of tumor compression of the inferior vena cava. Intraoperative ultrasonography was performed to determine the exact cutting line. After the hilar structures were dissected at mid-hilum, the proper hepatic artery was divided. Application of vascular clamps was followed by division on the portal vein and suprahepatic and infrahepatic vena cava. Right hepatectomy was performed with intermittent clamping (Pringle maneuver)[3], and hepatic parenchymal resection was performed with the Cavitational Ultrasonic Surgical Aspirator Excel CEM System (CUSA Excel CEMTM System; Valleylab, Boulder, CO, USA)[4]. Hepatic hemangiomas of the left lobe were not resected because the remnant liver would be reduced. After resection, a biliary leakage test was performed with injections of saline and air, and the leakage points were repaired with fine sutures. Hemostasis of the cut surface of the liver was achieved by ligation and the application of a fibrin glue spray (Bolheal; Chemo-Sero Therapeutic Research Institute, Kumamoto, Japan; Teijin Pharma Limited, Tokyo, Japan). An external drainage catheter (19-Fr. Blake Silicon Drain, Ethicon, Somerville, NJ, USA) was positioned at the site of resection. The weight of the resected specimen was 2,100 g. The operative time was 7 hours 58 minutes, and the blood loss was 3,035 g. Pathologic examination of the surgical specimen confirmed the presence of benign hepatic

Fig. 5 Computed tomography revealed hypertrophy of the left lobe of the liver after the operation.

hemangiomas. The postoperative course was uneventful, and the patient's appetite improved. The patient was discharged 8 days after the operation. Abdominal distension decreased, and laboratory data improved after the operation. Computed tomography revealed hypertrophy of the left lobe of the liver after the operation **(Fig. 5)**.

Discussion

Hemangiomas are the most common primary liver tumor, with an estimated prevalence in the general population of 0.4% to 7.3%[5,6]. Sixty percent to 80% of patients are women, most commonly aged 30 to 50 years, and tumors are rare in children[7]. Hepatic hemangiomas are usually solitary lesions, with multiple lesions present in only 10% of patients[7,8]. Some studies have suggested a possible relationship with the intake of steroid hormones[9,10] or female hormones[11], but such relationships are controversial[12].

Most hepatic hemangiomas remain stable in size[13,14]. A giant hemangioma, which is defined as a hemangioma larger than 4 cm in diameter, can cause symptoms and require treatment[15]. A hemangioma can cause congestion, bleeding, thrombosis, infarction, or Kasabach-Merritt syndrome and can spontaneously rupture. Through a mass effect, hemangiomas might show such symptoms as obstructive jaundice, biliary colic, and gastric outlet obstruction[1,15–21].

Kasabach-Merritt syndrome is a consumptive coagulopathy that was originally described in association with cutaneous hemangioma in children[22]. The primary pathophysiologic event of Kasabach-Merritt syndrome is clotting and fibrinolysis within the hemangioma. In the present case, laboratory data revealed pancytopenia and mild coaglopathy. Pancytopenia due to hypersplenism occasionally develops in patients with portal hypertension, even if splenomegaly is not detected. In our patient, the portal vein was compressed by the huge tumor, so hypersplenism due to portal hypertension and Kasabach-Merritt syndrome might have developed. Various treatments for pancytopenia due to portal hypertension have been reported[23,24]. In our patient, pancytopenia and mild coagulopathy resolved after the operation.

Indications for treatment, as proposed in the literature, include giant hemangiomas with progressive complications, an increase in size, and the possibility of malignancy[25]. Numerous treatments for hemangiomas have been described, such as drugs, ligation of the hepatic artery[26], selective transcatheter arterial embolization[27,28], radiation therapy[29], liver transplantation[30], and liver resection. However, except for liver transplantation and resection, these methods are unsuccessful in the long term. As a method of liver resection, enucleation should be considered for anterior and superficial hemangiomas[31,32]. Partial liver resection is indicated for larger hemangiomas in peripheral segments. Enucleation technique have been reported to cause no deaths and to have a low postoperative morbidity rate[33]. However, giant hemangiomas are usually hyperemic soft tumors with a thin, fragile wall, and hemostasis is difficult to establish once they start to bleed. The multiple resection planes in this case were anticipated to be large, nonanatomical, and to cause massive bleeding. In cases of giant or multiple hemangiomas, extended hemihepatectomy with Pringle's maneuver or total hepatic vascular exclusion to minimize blood loss is recommended.

The anterior approach for major right hepatectomy in which the right lobe is not mobilized was first described by Ozawa [34] as a "nonconventional approach" to advanced liver cancer

in an attempt to avoid prolonged rotation and displacement of the hepatic lobe. Rotation of the liver during hepatic resection can impair liver function. Liu et al.[35] have retrospectively compared the anterior approach with the conventional approach for major right hepatic resection to treat tumors of comparable size. The patients treated with the anterior approach had a significantly lower intraoperative blood loss and need for blood transfusion, a lower hospital death rate, a lower incidence of pulmonary metastases, and better disease-free survival and median overall cumulative survival. The liver-hanging maneuver, originally developed by Belghiti et al.[36], is a technique in which tape is passed between the anterior surface of the inferior vena cava and the liver, and the liver is suspended during the hepatic parenchymal transection in right hepatectomy. We selected the anterior approach for the present patient because mobilization of the right lobe was difficult, but liver-hanging maneuver could not be performed because the tumor had compressed the inferior vena cava. Right major hepatectomy for large primary liver cancers, tumor compression of the inferior vena cava, and the anterior approach without the liver-hanging maneuver are risks for massive bleeding during hepatectomy[37]. We were concerned about the possibility of uncontrolled bleeding during hepatic resection and of accidental injuries to major hepatic structures, including major Glissonian branches or hepatic veins compressed by giant tumor. We performed right hepatectomy carefully, but massive blood loss occurred.

In the present case, the patient's appetite and laboratory data improved after liver resection. Liver resection was a safe and effective treatment for giant hemangiomas causing abdominal symptoms.

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