Int Surg 2013;**98**:19–23

Case Report

Giant Hepatic Hemangioma Presenting as Gastric Outlet Obstruction

Cemalettin Aydin[1], Sami Akbulut[1], Koray Kutluturk[1], Aysegul Kahraman[2], Cuneyt Kayaalp[1], Sezai Yilmaz[1]

[1]*Liver Transplant Institute, and*

[2]*Department of Radiology, Inonu University Faculty of Medicine, Malatya, Turkey*

Hemangioma, a most frequently encountered primary benign tumor of the liver, is generally determined incidentally during the course of radiologic tests for other reasons. Most lesions are less than 3 cm and a significant proportion of patients are asymptomatic, although the size and location of the lesion in some patients may be associated with the onset of symptoms. Pressure on the stomach and duodenum of giant hemagiomas developing in the left lobe of the liver, in particular, may result in the development of abdominal pain, nausea, vomiting, and feeling bloated, which are characteristic of a gastric outlet obstruction. A 42-year-old man presented with findings of gastric outlet obstruction and weight loss as a result of a giant hepatic hemangioma.

Key words: Liver – Giant – Cavernous hemangioma – Gastric outlet obstruction – Enucleation

Hemangiomas, which are the most frequently encountered primary benign tumors in the liver, generally have a single subcapsular location, with most located in the posterior of the right lobe. Most lesions are less than 3 cm and a significant proportion of patients are asymptomatic.[1–4] In symptomatic patients, symptoms are associated with the size and location of the lesion, such as nausea, vomiting, abdominal pain, palpable mass, obstructive jaundice, gastric outlet obstruction and hemorrhage, consumption coagulopathy, and thrombocytopenia, which are characteristic of Kasabach-Merritt syndrome.[1,2,5] Although there is no general consensus on size, hemangioma >4 cm are referred to as giant.[1,2,5–8] In asymptomatic patients, although there is agreement that there should be close monitoring rather than any treatment, at present there has been no consensus as to whether medical treatment, interventional radiologic treatment, or surgical treatment should be applied to symptomatic cases. This study aimed to present an approach to a case of giant hepatic hemangioma that developed in the left lobe of the liver with findings of gastric outlet obstruction.

Reprint requests: Sami Akbulut, MD, Liver Transplantation Institute, Inonu University Faculty of Medicine, 44280, Malatya, Turkey. Tel.: +90 422 3410660; Fax: +90 422 3410036; E-mail: akbulutsami@gmail.com

Case Report

A 42-year-old male patient presented at an external health center with complaints of nausea, projectile vomiting, and abdominal pain that had started 2 weeks previously and worsened in the past 3 days. The patient reported that these complaints had started a long time previously and in the past 2 months he had lost at least 10 kg from loss of appetite. The physical examination revealed a positive Murphy's sign and a palpable mass in the right upper quadrant extending to the epigastrium. Abdominal ultrasonography determined that the gallbladder was hydropic and the wall thickness was significantly increased. Blood cell count revealed leukocytosis at 14.500/mm^3 (normal range, 4–11.000/mm^3). When the findings of the clinical, physical, and radiologic examinations were considered, a diagnosis of acute cholecystitis was made. Laparotomy determined a large mass completely covering the left lobe of the liver that was causing severe compression of the stomach both laterally and inferiorally. Cholecystectomy was performed because of hydropic gallbladder. After the cholecystectomy, the patient was referred to our center for further tests. The blood tests done in our clinic determined measurements of hemoglobin 12.6 g/dL, platelet 354,000/mL, international normalized ratio (INR) 1.36, albumin 3 g/dL, total/direct bilirubin 0.93/0.39 mg/dL, aspartate aminotransferase 15 U/L, alanine aminotransferase 35 U/L, and alkaline phosphatase (ALP) 257 U/L. Hepatitis markers and other tumor markers were negative. With the contrast-enhanced multislice computed tomography (CT), a lesion was observed 25 × 16 × 16 cm, completely filling the left lobe of the liver, showing exophytic extension and pressing toward the posterior-lateral of the stomach. In the CT slices without contrast, the lesion was observed to cover the lobe contours (giant cavernous hemangioma) (Fig. 1). Because the mass was compressing the middle hepatic vein, hypodense areas were seen, consistent with congestion of the neighboring segments 5 and 8. Surgery was undertaken because of the pressure being put on both the gastrointestinal system and the middle hepatic vein. When laparotomy performed, a great number of adhesions were seen in the hilar area because of previous surgical procedures. The hepatoduodenal ligament was turned for the Pringle maneuver. Then the demarcation line was defined by suspending the left hepatic artery and the left portal vein. The left edge of the mass was very close to the demarcation line and the mass was seen to be touching the middle hepatic vein. The surgery was started from the anterior edge of the liver and continued to the posterior edge, protecting the middle hapatic vein wall. Finally, the mass was removed by enucleation as to protect the middle hepatic vein. Therefore, during this procedure segments 2, 3, and 4 were resected. On the cholangiography taken after enucleation, the bile ducts were seen to be intact (Fig. 2). The surgical procedure took approximately 3 hours. Intraoperatively there was blood loss of 750 to 800 mL and 2 units of erythrocyte replacement were administered. The postoperative Doppler ultrasonography showed good flow in the venous drainage and hepatic artery. No complications developed in the postoperative period.

Discussion

Hemangioma are benign harmartomatous tumors that develop as the result of the rapid proliferation of vascular endothelial cells over time.[9] Although most frequently seen on the skin of the head and neck, hemangiomas can be seen on nearly every surface and organ of the body. They are most often seen in the liver.[9–11] The incidence of hemangiomas varies from 0.4% to 20%.[1–3,6] Hemangioma are often seen between the ages of 30 and 50 years and 5 to 6 times more often in women than in men.[4,6,7,9,10] Therefore, it has been suggested that there may be a relationship between the disease and the use of exogenous steroids or female-specific hormones, although the relationship has not been clearly shown.[2,3,10]

Because most hepatic hemangioma are asymptomatic, they are often recognized incidentally during radiologic tests for other reasons. However, in some patients the location and size of the lesion can cause various signs and findings. According to a previous study,[4] although 40% of hemangioma about 4 cm in diameter are symptomatic, 90% of those approximately 10 cm in diameter are symptomatic. The most frequently seen symptom is pain in the right upper quadrant. Pain results from the stretching of the Glisson capsule, the development of thrombus within the hemangioma, increased pulsating blood flow toward the lesion, and the development of necrosis in the lesion.[10]

Gastric outlet obstruction findings, such as nausea, vomiting, and bloating, result from pressure on the stomach and the duodenum by giant hemangioma.[2,7] Congestion of various liver segments, renal insufficiency, edema of the lower

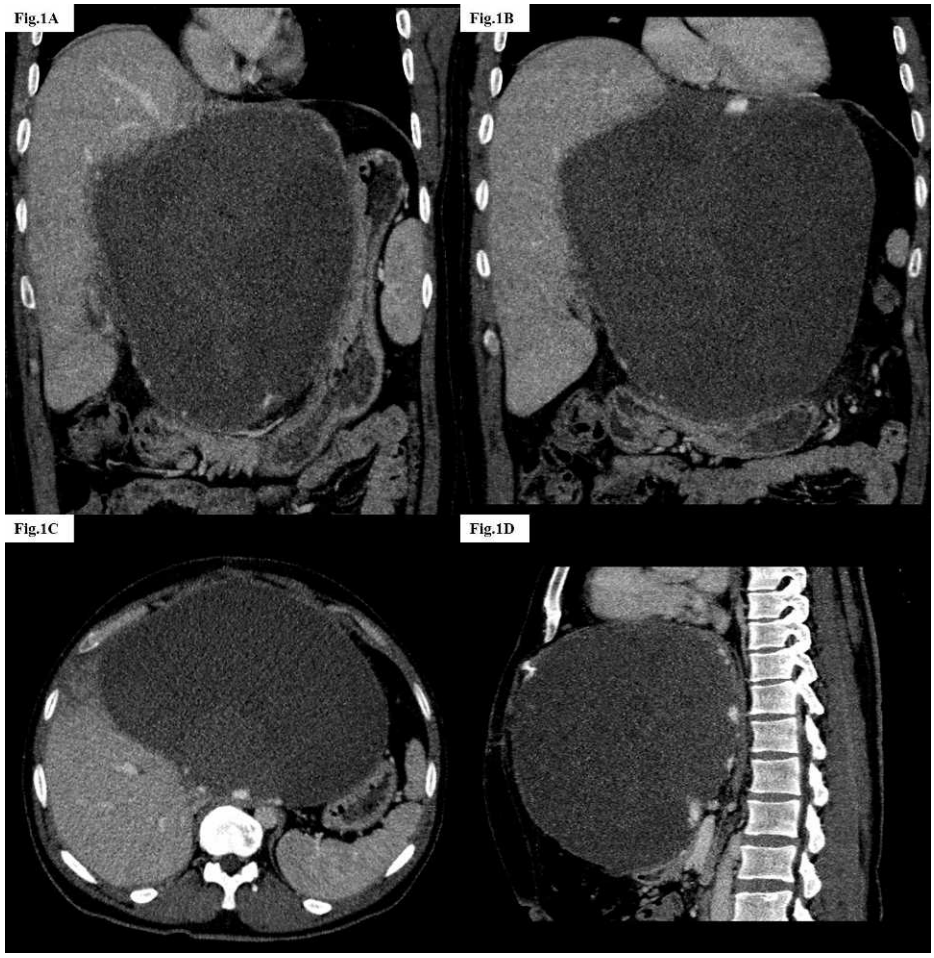

Fig. 1 In the multislice computed tomography (CT) images taken in the portal venous phase, coronal (A), coronal (B), axial (C), and sagittal (D), a mass is observed consistent with a giant cavernous hemangioma, filling the left lobe of the liver, showing exophytic extension, compressing adjacent structures, and showing peripheral intermittent nodular contrast enhancement.

extremities, and clinical findings of Budd-Chiari syndrome may develop due to the compressive effect of hemangiomas on the hepatic vein and the inferior vena cava.[3] In addition, giant hemangioma may cause congestive heart failure resulting from arteriovenous shunt, and this is associated with Kasabach-Merritt syndrome characterized by thrombocytopenia, microangiopathic hemolytic anemia, and consumptive coagulopathy.[4,5]

Diagnosis of hepatic hemangioma can be made easily with radiologic diagnostic tools such as ultrasound, CT, magnetic resonance imaging, and single-photon emission CT, either alone or in combination.[11,12] Although angiography is the gold standard in the diagnosis of hemangioma, its greatest disadvantage is that it is an invasive procedure. However, it is a method that can be used in cases where differential diagnosis of hepatic hemangioma from other malignant lesions could not be made by other radiologic tests.[5] In cases where a diagnosis cannot be made by any radiologic means, differential diagnosis can be achieved by needle biopsy, although there is a risk of hemorrhage. However, in cases where there is a suspicion of malignancy, it should not be forgotten that tumor inoculation can be made along the biopsy tract.[11]

The most appropriate approach for asymptomatic patients is close monitoring rather than any form of treatment.[3,6,7] Spontaneous or traumatic rupture, intratumoral hemorrhage, consumptive coagulopathy, and rapid growth of the tumor are all definite indications for surgical treatment. Persistent abdominal pain, obstructive jaundice, portal hypertension, other compression symptoms (*e.g.*, gastric outlet obstruction), tumor diameter >5 cm, superficial

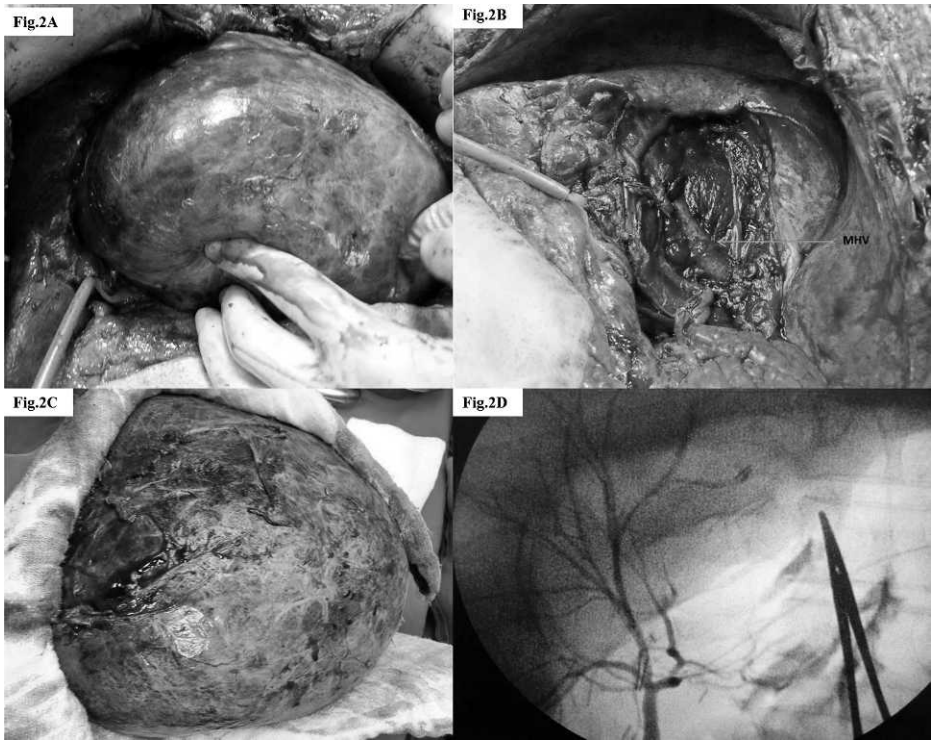

Fig. 2 Images obtained at different stages of surgery. (A) Image of the mass completely filling the left lobe. (B) Image showing preservation of the middle hepatic vein after resection of the mass in the left lobe. (C) Image of the resected mass. (D) Cholangiography image taken to evaluate the bile ducts after resection.

location, and suspicion of malignancy are indications for surgical treatment.[2,7,11]

The most frequently used methods of surgical treatment are hepatic artery ligation, enucleation, resection, and occasionally liver transplantation.[2,4,7] In addition, some studies have shown positive results from transartery embolization, liver radiotherapy, systemic corticosteroid treatment, and interferon treatment.[4] For patients with indications for surgical treatment, there is no clear consensus as to which surgical technique should be used. In addition to the proponents of partial resection, more surgeons favor enucleation, which is thought to be a safer method.

With the advent of enucleation in 1988, many surgeons began to advocate the enucleation approach for hepatic hemangioma. The dissection border of this technique is where fibrous cleavage forms between the normal liver tissue and the capsule of the hemangioma. Therefore, in the safe advancement of this plan, the procedure can be made without damaging either the bile ducts or the major hepatic vascular structures. Previous nonrandomized studies have shown that enucleation is safer, quicker, has less blood loss, and lower rates of morbidity and mortality than resection. However, due to risk of massive blood loss with lesions >10 cm in diameter or lesions with a central location, enucleation may not be an appropriate choice.[2,13] We believe that enucleation can be applied successfully to large or centrally located lesions, when the Pringle maneuver is used at appropriate intervals.[14] Partial liver resection is more effective for large lesions located in peripheral segments. The most suitable approach for large or multiple hemangioma is to use the Pringle maneuver or total hepatic vascular exclusion together with extended hemihepatectomy.[6,7] In unresectable lesions, when there is multiple bilobar involvement and in symptomatic cases with hepatic hilum involvement, liver transplantation is the most appropriate approach.[4,6] After resection in multiple and bilaterally located hemangioma, it is necessary to take the liver reserve into consideration. In these patients, a suitable approach is to first excise the lesion that is causing the symptoms. When all of these treatment modalities are examined, apart from extended resection and transplantation, long-term results have not been good.[3]

References

1. Koszka AJ, Ferreira FG, de Aquino CG, Ribeiro MA, Gallo AS, Aranzana EM *et al*. Resection of a rapid-growing 40-cm giant liver hemangioma. *World J Hepatol* 2010;**2**(7):292–294
2. Uetama T, Yoshida H, Hirakata A, Yokoyama T, Maruyama H, Suzuki S *et al*. A symptomatic giant hepatic hemangioma treated with hepatectomy. *J Nihon Med Sch* 2011;**78**(1):34–39
3. Jiang H, Chen Z, Prasoon P, Wu H, Zeng Y. Surgical management for giant liver hemangiomas greater than 20 cm in size. *Gut Liver* 2011;**5**(2):228–233
4. Vagefi PA, Klein I, Gelb B, Hameed B, Moff SL, Simko JP *et al*. Emergent orthotopic liver transplantation for hemorrhage from a giant cavernous hepatic hemangioma: case report and review. *J Gastrointest Surg* 2011;**15**(1):209–214
5. Moon HK, Kim HS, Heo GM, Shin WG, Kim KH, Jang MK *et al*. A case of pedunculated hepatic hemangioma mimicking submucosal tumor of the stomach. *Korean J Hepatol* 2011;**17**(1): 66–70
6. Sood D, Mohan N, Singh A, Buxi TB, Nundy S, Soin AS. Living donor liver transplantation for giant cavernous hemangioma of liver in a child. *Pediatr Transplant* 2011;**15**(7):E135–138
7. Choi J, Lee YJ, Hwang DW, Chon SH, Nagpal A, Park KM. Surgical treatment of giant hepatic hemangiomas: technical point of view. *Am Surg* 2011;**77**(1):48–54
8. Jain V, Ramachandran V, Garg R, Pal S, Gamanagatti SR, Srivastava DN. Spontaneous rupture of a giant hepatic hemangioma—sequential management with transcatheter arterial embolization and resection. *Saudi J Gastroenterol* 2010; **16**(2):116–119
9. Van Malenstein H, Maleux G, Monbaliu D, Verslype C, Komuta M, Roskams T *et al*. Giant liver hemangioma: the role of female sex hormones and treatment. *Eur J Gastroenterol Hepatol* 2011;**23**(5):438–443
10. Duxbury MS, Garden OJ. Giant haemangioma of the liver: observation or resection? *Dig Surg* 2010;**27**(1):7–11
11. Corigliano N, Mercantini P, Amodio PM, Balducci G, Caterino S, Ramacciato G *et al*. Hemoperitoneum from a spontaneous rupture of a giant hemangioma of the liver: report of a case. *Surg Today* 2003;**33**(6):459–463
12. Unal E, Francis F, Aquino A, Xu R, Morgan G, Teperman L. Liver transplant for mixed capillary-cavernous hemangioma masquerading as hepatocellular carcinoma in a patient with hepatocellular carcinoma. *Exp Clin Transplant* 2011;**9**(5):344–348
13. Xia F, Lau WY, Qian C, Wang S, Ma K, Bie P. Surgical treatment of giant liver hemangiomas: enucleation with continuous occlusion of hepatic artery proper and intermittent Pringle maneuver. *World J Surg* 2010;**34**(9):2162–2167
14. Fu XH, Lai EC, Yao XP, Chu KJ, Cheng SQ, Shen F *et al*. Enucleation of liver hemangiomas: is there a difference in surgical outcomes for centrally or peripherally located lesions? *Am J Surg* 2009;**198**(2):184–187