

Early Treatment for Symptomatic Giant Hepatic Hemangioma: Report of Three Cases and Literature Review

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Abstract: Hemangiomas are the most common benign tumors of the liver. Most of them remain stable over time. Fewer than 10% of cases undergo enlargement and only some of them cause symptoms. When a hemangioma keeps growing and become symptomatic, it can be managed by surgery, Transcatheter Arterial Embolization (TAE) and Radiofrequency Ablation (RFA). In our experience, we suggest early intervention without delay and we preferred surgical resection. We report three cases of symptomatic giant hemangioma of different sizes (6, 11 and 22 cm, respectively). They had symptoms included abdominal distention, pain, poor appetite, bilateral legs pitting edema and one of them presented as intractable hiccup and was misdiagnosed as gastrointestinal reflux. All of them were treated by surgery and their symptoms were all relieved. Their postoperative courses were uneventful. In two of our cases, they had symptoms more than one year and their tumors grow up over 10 cm with compression on the inferior vena cava tightly. Early surgical resections in symptomatic giant hepatic hemangiomas may make operation more safely in cases without marked surgical contraindication and not high risks patients. In this report, we also review the literatures about the treatment of symptomatic hepatic hemangiomas.

Key words: Giant hepatic hemangioma, hepatectomy, transarterial embolization, radio frequency ablation

INTRODUCTION

Hemangiomas are congenital vascular malformations and are the most common benign tumors of the liver. A symptomatic hemangioma is not so common. For the majority, it is often asymptomatic and does not growth up. Only fewer than 10% of cases undergoes enlargement (Griffa *et al.*, 2005) and some of them become huge enough to cause symptom. The hemangioma is a benign liver tumor. How to manage a symptomatic hemangioma? Several methods including surgery, transarterial embolization and radiofrequency ablation have been reported.

Here we present three symptomatic patients that their symptoms bothered them so much. Although hemangioma was benign, the symptoms has interfered their quality of life significantly. They all were treated surgically.

CASE REPORT

Case 1: A 40-year-old woman was previously healthy. A hepatic hemangioma was incidentally found in a physical check up by ultrasonography about 8 years ago. At that time, the tumor size was only 3-4 cm and it kept growing gradually, about 1cm in diameter ever year. However, she

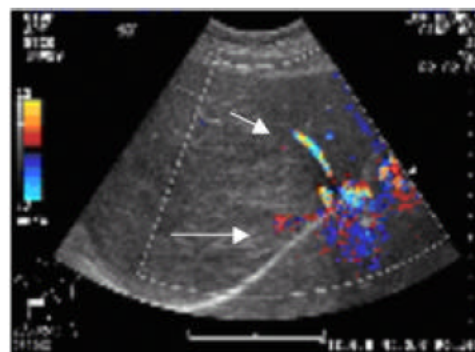


Fig. 1: Doppler sonography: A hyper-echoic mass in the right lobe of the liver. The right hepatic vein was merged in the mass (long arrow). The middle hepatic vein was very close to the mass (short arrow)

did not have any symptoms for eight years. When the tumor size reached 11×8 cm, she felt intermittent pain on right upper quadrant abdomen and right back. She also felt fullness and early satiety easily. She was treated symptomatically for one year. As the persistent symptoms bothered her physically and psychologically, she came to our hospital for further management.



Fig. 2: A huge mass with peripheral enhanced nodularity nearly occupied the whole segments 7 and 8 and extended to the segments 5 and 6. The IVC was compressed to the medial aspect by the mass. The right hepatic vein cannot be seen clearly here. The middle hepatic vein was very close to the mass (arrow)

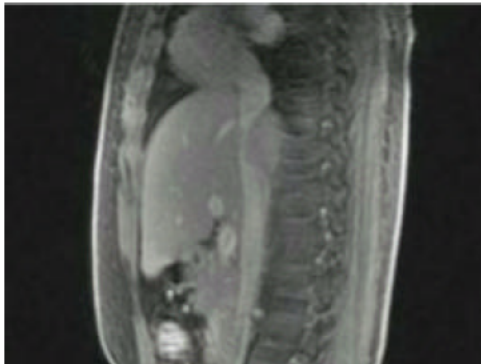


Fig. 3: MRI sagittal view revealed the location of the tumor and the IVC. The tumor was abutting to the IVC with extension to the posterior aspect of the IVC and near the orifice of the hepatic vein

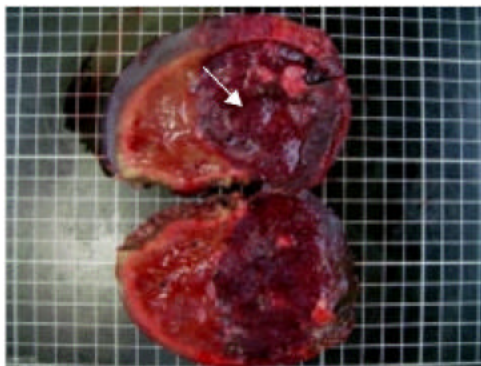


Fig. 4: Cut section of liver: the reddish tumor (arrow) measures 11.5x8x7 cm in size

On physical examination, she had a mild distended abdomen with slight tenderness on right upper quadrant. Bilateral lower extremities were free from edema. No definite jaundice, no pallor. The laboratory data revealed normal CBC and normal liver function test. Hepatitis B Virus (HBV) and Hepatitis C Virus (HCV) markers were negative and α Fetal Protein (AFP) level was 14.56 ng dL^{-1} (normal value: 0~10).

Her sonography (Fig. 1) revealed a hyper-echoic mass. On Doppler sonography, it showed that the right hepatic vein was merged in the mass. The middle hepatic vein was very close to the mass.

Her Computed Tomography (CT) scan (Fig. 2) showed a huge hypodense mass nearly occupied the whole segments 7 and 8 and extended to the segments 5 and 6. Her CT and Magnetic Resonance Imaging (MRI) (Fig. 3) showed compression of the tumor on the intrahepatic IVC near the orifice of the hepatic vein and extended to the posterior aspect of the IVC.

She received right hepatectomy and cholecystectomy (Fig. 4). The tumor presented as the image studies that compressed on the IVC tightly. It cannot be exposed from the lateral approach. The postoperative course was uneventful.

Case 2: This 48 years old man has similar clinical presentation and symptoms as those mentioned in the first case. He had abdominal distention, tenderness, fullness and poor appetite for more than one year. His tumor was so huge that on physical examination it revealed a protruded right upper abdomen and a palpable liver with mild tenderness. Pitting edema was noted in bilateral lower limbs. His abdominal CT scan (Fig. 5) showed a huge hemangioma $22.5 \times 20 \times 7 \text{ cm}$ occupying the whole right lobe of liver and extending to segment 4. His lateral segment was hypertrophic. The tumor compressed and encased the IVC tightly.

He received extended right hepatectomy by anterior approach without mobilization of the liver (Fig. 6). The pitting edema of lower limbs subsided after the surgery. His post-operative course was uneventful.

Case 3: A 36/o woman, previously healthy, got hiccup for 7 months. The frequency was about 4 times in every minute. It was also accompanied with nausea and vomiting, especially after a meal. The symptoms became more significant after exercise. The hiccup could not stop excepted she went into sleep. She also has received further evaluation and the sonography show a mass in the liver. The CT scan (Fig. 7) revealed a hemangioma in the liver surface of posterior aspect of segment 7 and it attached on the diaphragm. She did not have psychological problem and was treated as gastroesophageal reflux with medication for 5 months. However, the hiccup persisted and was still intractable.



Fig. 5: A huge peripheral enhanced tumor in the right lobe of liver and extends to segment 4 with IVC compression significantly

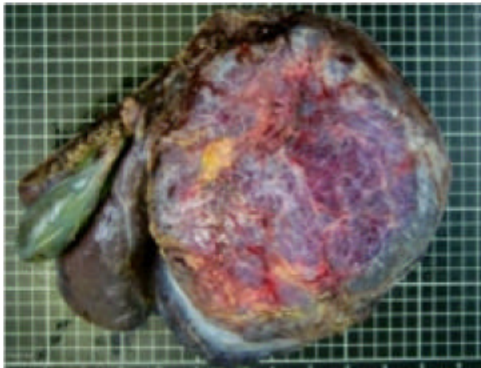


Fig. 6: View from posterior aspect: A huge tumor measuring 22.5×20×7 cm nearly occupies whole right lobe with an intact capsule and smooth surface

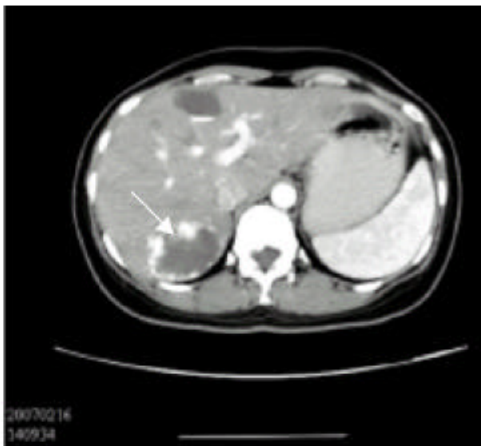


Fig 7: A 6 cm peripheral enhanced hypodense mass at dome in S7 (arrow) of right hepatic lobe. A cyst is seen on S8

The persisted hiccup bothered to her daily life and work extensively. She was considered to undergo the surgery for hemangioma due to the possible etiology of the hiccup.

We performed a segmental hepatic resection to remove the hemangioma at the dome of right lobe liver. After the surgery, her hiccup stop dramatically. Her post-operative course was smoothly. The hiccup did not recurrence in the recent follow up after 5 months of operation.

DISCUSSION

A giant hemangioma is considered when its size is more than 4 cm. Most of them remain stable and no symptom over time. But some of them may become symptomatic. The most common symptom was abdominal pain or discomfort.

The hemangioma may ruptures spontaneously, or with minimal trauma and present as hemoperitoneum. A few case reports have been reported, especially when the tumor is close to surface. The incidence is about 1~4% and has been described in about 32 cases in the literature with a high mortality (36~69%) (Griffa *et al.*, 2005; Corigliano *et al.*, 2003; Cappellani *et al.*, 2000).

It is rare that large hemangiomas sequester and destroy platelets, causing consumptive coagulopathy, symptomatic thrombocytopenia, known as Kasabach-Merritt syndrome (Courtney *et al.*, 2004; Cui *et al.*, 2003).

The indications for resection is rupture, rapid increase in size, development of the Kasabach-Merritt syndrome and to make a definitive diagnosis in equivocal cases (Corigliano *et al.*, 2003). Liver transplantation, sometimes may be the choice of treatment for giant hemangioma as kasabach-Merritt syndrome developed (Kumashiro *et al.*, 2002; Ferraz *et al.*, 2004).

For the symptomatic patients, it is advocated that they should undergo a thorough evaluation to find out any other cause for the symptoms. If the hemangiomas are the reasons to induce pain, these patients are candidates for resection (Courtney *et al.*, 2004).

When talking about surgery, it still should be emphasized that the size of lesion is not a criterion for resection during management of giant liver haemangioma. Patients who suffered persistent significant symptoms are the candidates for resection. Just like the cases we present here, the symptoms are not directly related to the size of the tumor. The smaller giant hemangioma also can cause worrisome symptoms as intractable hiccup in the third case. In Yoon *et al.* (2003) study, among patients with symptom before resection, 96% had resolution of symptoms after

operation (Yoon *et al.*, 2003). Few patients have post-operative wound pain but for the majority of patients, surgical resection can achieve the best result of symptom relief. For the smaller tumor, enucleation is preferred. If the tumor location precludes safe enucleation or the tumor is huge and occupies the right (left) lobe, even compressing the major vessels as the cases we present, for the patient's safety, anatomic resections is the procedure of choice (Demircan *et al.*, 2005). In patient without cirrhotic change, the liver parenchyma is healthy and it is safer and easier to get meticulous hemostasis during resection than a cirrhotic liver. But it should be based on the balance between the benefits from surgical resection and the estimated operative risk of patients (Corigliano *et al.*, 2003). In a huge hemangioma, as in the second case, that may preclude the safety and fully mobilization of the liver, anterior approach hepatectomy can be applied. The operative morbidity and mortality are low in experience hands

Besides surgical resection, there are other modalities available to manage symptomatic hemangioma in the literature. Transcatheter arterial embolization and radiofrequency ablation, just like the treatment modalities for malignancy, they can be applied to manage hepatic hemangioma, especially for the high surgical risk patients.

There were several reports from China and India that TAE decreases tumor sizes and results in symptom relief in most reports. In Zeng *et al.* (2004) report, a series of 98 patients underwent TAE. Tumor diameters decreased from 9.7 +/- 2.3-5.6 +/- 1.6 cm 6 months after the treatment. The clinical symptoms were relieved in all 53 symptomatic patients. But transient impairment of liver function in 77 cases was noted. Zeng *et al.* (2001) But the paper lacked the long term outcome of these patients.

There were also some drawbacks with this technique. In Huang *et al.* (2002) report, six cases of severe biliary and hepatic complications including liver abscess, gallbladder necrosis, gangrene and bile duct stricture after hepatic artery embolization for hepatic hemangioma has been reported (Huang *et al.*, 2002). The tumor size of these patient ranged from 3-6cm in diameter. His conclusion is that hepatic arterial embolization of hemangioma may result in severe destructive biliary damage and its indiscriminal use should be prohibited. We have no experience of TAE for hemangioma at our hospital and there is also very limited literature report in Taiwan. In the literature, there is also no long term follow up result of the size after TAE of hemangioma.

But in case of rupture, TAE is considered to be a valid procedure in stanching or reducing the hemorrhage, thus making it a safer hepatic resection (Adam *et al.*, 1970). Suzuki *et al.* (1997) suggested the use of preoperative TAE in patients with consumption

coagulopathy related to intravascular coagulation in the hemangioma (Suzuki *et al.*, 1997).

Ultrasonography-guide percutaneous Radio Frequency Ablation (RFA) is another non-operative treatment. In Cui *et al.* (2003) s series, 12 patients with 15 hepatic cavernous hemangiomas (2.5-9.5 cm) were treated with RFA. The ablated lesions were shrunk and the shrunk range was 38-79% (Yan *et al.*, 2003). For tumor too close to the hilum and the liver boarder, it should be careful to avoid major complication such as bleeding, biliary complication and hollow organ perforation. But it also did not provide long term follow up result of these patients.

These non-resection modality can be applied in selected patients, especially for those with smaller symptomatic tumor and those are high risk for operation.

Intervention is needed when the tumor start to cause symptoms. In our two cases, they have symptoms more than one year. If they were managed earlier, tumor sizes might not so huge that make operation so risky, especially during exposure of IVC. In the first case, the IVC could not be exposed from lateral approach safely, in the second case, liver could not be mobilized and anterior approach was done.

For huge symptomatic giant hemangiomas, two steps management by combining both non-surgical and surgical modalities may reduce the size and make operation easier. But there is no experience about this in the literature.

In two of our patients, the IVC was compressed markedly, one even present lower limbs pitting edema, but fortunately there were no definite thrombus formation at the stenotic area of IVC at the time of operation. A case of pulmonary embolism due to compression of IVC by a hepatic hemangioma was reported by Paolillo *et al.* (1993). It is also a catastrophe to relieve the thrombus during the resection of tumor that leads to pulmonary embolism.

We do not know the tumors in our first and second cases would keep growing to occupy the whole liver or not. It has been reported that a giant hemangioma extended entire liver and result in respiratory restriction and compression of IVC, a low platelet count and DIC. Finally, liver transplantation was performed (Kumashiro *et al.*, 2002; Ferraz *et al.*, 2004).

For the third case, we may argue whether the hiccup is really related to the hemangioma or not because the tumor capsule is intact and not invasion to diaphragm was seen. The tumors in our two other cases also located in the dome, but they did not have hiccup. The hemangioma was found when she started hiccupping but she was managed medically as gastrointestinal reflux. In the literature review, hiccup induced by a hemangioma has not been reported. It was diagnosed as the most possible etiology to cause hiccup in this patient by exclusion of other causes because her intractable hiccup for long duration disappeared dramatically after the operation.

CONCLUSION

Fewer than 10% of cases undergo enlargement (Griffa *et al.*, 2005) and only some of them cause symptoms. Therefore, intervention is not necessary in the majority of stable or non-symptomatic growing hemangioma. In the contrary, for a symptomatic patient, either early surgical or non-surgical modality is needed. But the long-term results of TAE and RFA have not been reported yet. Besides, non-surgical modalities have more limitation in practical use in huge tumor or tumor at peripheral location. Surgery can definitely relieve the symptom. Thus, for growing and symptomatic hemangiomas, we suggest early intervention and preferred early surgical resection in patients without definite surgical contraindication and not high risk patients.

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