

Case Report

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Surgical Treatment of Giant Hepatic Hemangioma in a Patient with Congenital Thoracic Scoliosis: A Case Report

Thuan Vo Duy* and Song Huy Nguyen-Dinh

Liver Tumor Department, Oncology Center, Cho Ray Hospital, HCMC, Vietnam

ABSTRACT

Background: Hepatic Hemangioma (HH) is the most common benign primary hepatic neoplasm, which is usually asymptomatic and does not require intervention. The definition of a Giant HH is an HH with a size of more than 5 cm and symptomatic abdominal pain due to compression of adjacent organs. In this study, we report a rare case of giant HH in a young woman with congenital thoracic scoliosis, successfully treated via primary surgical resection guided by three-dimensional (3D) simulation.

Case Report: We present the case of a 30-year-old woman who complained of various gastrointestinal symptoms occurring 4 months after the delivery of her second child. A series of examinations was done, which revealed two giant hemangiomas. The patient was planned for surgery, and the tumors were successfully resected. The patient was discharged after eight days of hospitalization without any severe complications.

Conclusions: Giant HH patient with congenital thoracic scoliosis has not been reported in previous studies, we suggest that patients with giant HH should undergo surgical treatment when the diagnosis is made to reduce symptoms and prevent the risk of traumatic or iatrogenic rupture and this operation is not affected by her thoracic scoliosis. But we should make a detailed plan before surgery to performed liver resection.

*Corresponding author

Thuan Vo Duy, Liver Tumor Department, Oncology Center, Cho Ray Hospital, HCMC, Vietnam.

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Introduction

Hepatic hemangioma (HH) was initially described in the English literature in a translation of Frerich's "A Clinical Treatise on Disease of the Liver" in 1861 [1]. That is a vascular tumor, accounting for over 73% of benign liver neoplasms. In autopsy, the rate of HH accounts for up to 7.3% [2]. In general, HH is classified into three types: cavernous, capillary, and sclerotic HH. Capillary HH are usually less than 3cm in size, whereas cavernous HH are usually larger than 5cm, and sclerotic HH are characterized by fibrous connective tissue in the tumor [3]. Microscopically, HH is composed of multiple thick-walled abnormal blood sinuses of different sizes. The luminal wall is lined with endothelial cells [4]. While HHs can occur at any age, they are most frequently diagnosed between ages 30 and 50 and are more common in women [4]. Giant HHs are generally defined as lesions more than 5 cm, but the final consensus has yet to be reached. Ketchum used the terminology "Mega" for hepatic hemangiomas measuring larger than 10 cm [2]. Another author applied enormous hemangiomas as those more than 15 cm in size [5]. Hormonal changes during pregnancy may trigger rapid lesion growth, and HHs are often incidentally discovered via ultrasound or MRI. Giant HH may present with nonspecific symptoms such as abdominal pain, loss of appetite, fatigue, bloating, and compression of adjacent organs, causing nausea and vomiting. On physical examination, the lesion could also be palpable on the abdominal wall [4].

Diagnosis of HH is generally based on imaging findings. The result of abdominal ultrasound shows a homogeneous hyperechoic mass (small tumor) or an inhomogeneous hyperechoic mass (large tumor) in common. On abdominal computed tomography scans with contrast, the tumor often enhances the rim in the arterial phase and gradually fills with contrast in the venous and late phases. On abdominal MRI images with contrast, the tumor has the characteristics of no hyperintensity at the T1 phase, hyperintensity at the T2 phase, hyperintensity at the contrast injection phase, and gradually increasing contrast agents within the tumor in different areas. Biopsy is rarely used for diagnosis due to its high risk of bleeding. Giant HH can cause severe complications of intra-tumoral bleeding, like Kassabach-Merrit syndrome (combined giant Hemangioma with thrombocytopenia syndrome) or rupture [4].

There are various treatments for HH, such as surgical resection, transarterial embolization (TAE), radiofrequency ablation (RFA), microwave ablation (MWA), immunotherapy, and systemic chemotherapy, but there are still no best recommendations to treat giant HH. In clinical practice, surgical resection has been shown as an effective intervention and has been widely accepted in cases of large symptomatic HH or tumor rupture.

Case Report

A 30-year-old female was admitted to the Department of Liver Tumor, Cancer Center, Cho Ray Hospital, Ho Chi Minh City, Viet Nam) for surgery after four months of diagnosis of HH at time of her delivering with uncomfortable symptoms including

fatigue, discomfort, fullness, abdominal distention, palpable mass in epigastrium or subcostal region. Her medical history was recorded with a congenital thoracic scoliosis. She was 1.35m height, 40kg weight, and BSA was 1.5.

The laboratory findings on admission were as follow: Complete blood count revealed red blood cell 2.98 T/L (3.8-5.5T/L), hemoglobin 8.8 g/dL (13.0- 18.0 g/dL), white blood cell 6.85/mm³ (4.0-11.0/mm³), and platelet 159/mm³ (200-400/mm³), prothrombine time 23.5 seconds (12-14 seconds), INR 1.3. Blood chemistry showed total bilirubin 0.6 mg/dL (0.2–1 mg/dL), direct bilirubin 0.15 mg/dL (0.0–0.2 mg/dL), AST 98.9 U/L (9–48 U/L), ALT 97 U/L (5–49 U/L), BUN 19.3 mg/dL (7–20 mg/dL), creatinine 0.73 mg/dL (0.7–1.5 mg/dL), ferritin (i2000) 20.42 ng/ml (Male: 20-400 ng/ml; female: 6-180 ng/ml). Serological markers of hepatitis B and C were negative. The levels of tumor markers, including alpha-fetoprotein, carbohydrate antigen 19-9, cancer antigen 12.5, and carcinoembryonic antigen, were within the normal range.

The patient was also administered a transfusion with 1 unit of red blood cells (350ml) and 5 units (200ml/unit) of frozen fresh plasma

before surgery. The abdominal US showed many hyperechoic and inhomogeneous lesions in the liver, and Computed tomography (CT) with contrast revealed multiple tumors located on the left and right liver segments of the liver with peripheral enhancement in the arterial, gradual central filling in portal venous, and delayed phase (Figure 1).

On abdominal CT scan with contrast, the two biggest lesions were about 18 cm and 20 cm in diameter, located in the 5th-6th liver segment and in the left liver; some small remaining tumors were about 1-2cm. A diagnosis of multiple hemangiomas was confirmed. We used the 3D simulation system of Platform Marian software for finding the relationship between tumor and intrahepatic vessels, and the prediction of future remnant liver volume more clearly (Figure 2a, 2b). This system showed that the healthy liver volume was 923 cm³, and the hepatic lesion volume was 2635 cm³ [3]. A left hepatectomy and a non-anatomical resection of hemangioma located in the 5th-6th liver segment was indicated, with a prediction of residual liver about 648 cm³ (61%) when compared to the standard liver volume.

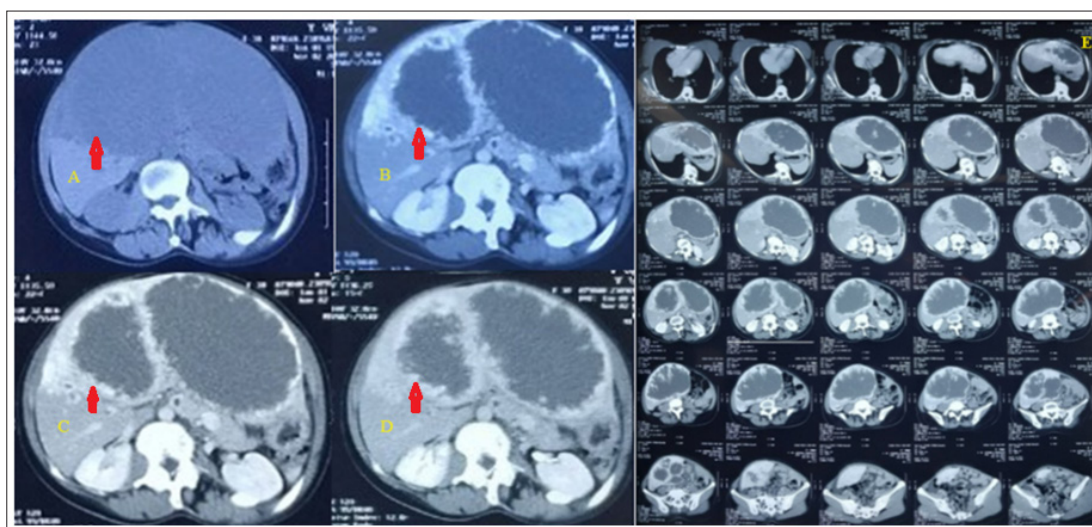


Figure 1: Abdominal Computed Tomography Showing: (A) Hypodense lesion in the non-contrast phase, (B) Peripheral enhancement in arterial phase, (C) Central filling in venous phase and (D) delayed phase. (E) Images of hemangiomas in the venous phase.

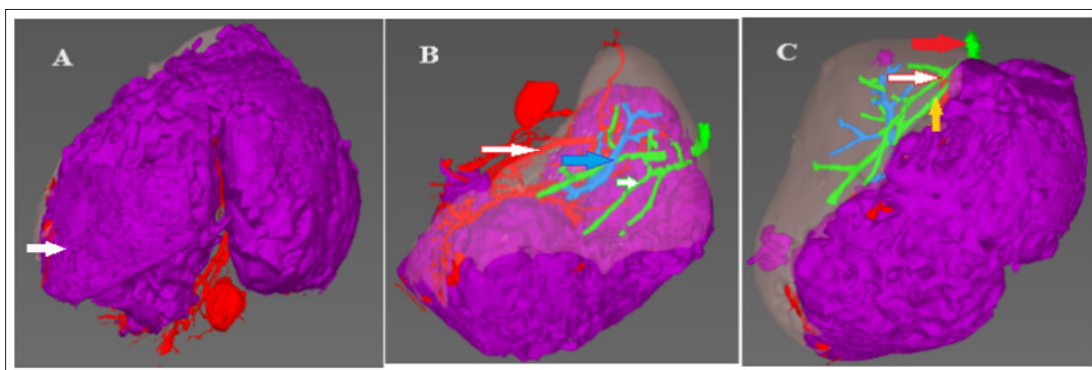


Figure 2a: 3D Simulation System of Platform Marian Software before Surgery

A (Anterior surface): Hemangioma (white arrow); B (Posterior surface): branch of right hepatic artery (red outlined arrow), right portal vein (blue arrow), hepatic vein (green outlined arrow); C. Inferior Vena Cava (red arrow), right hepatic vein (white arrow), middle hepatic vein (yellow arrow).

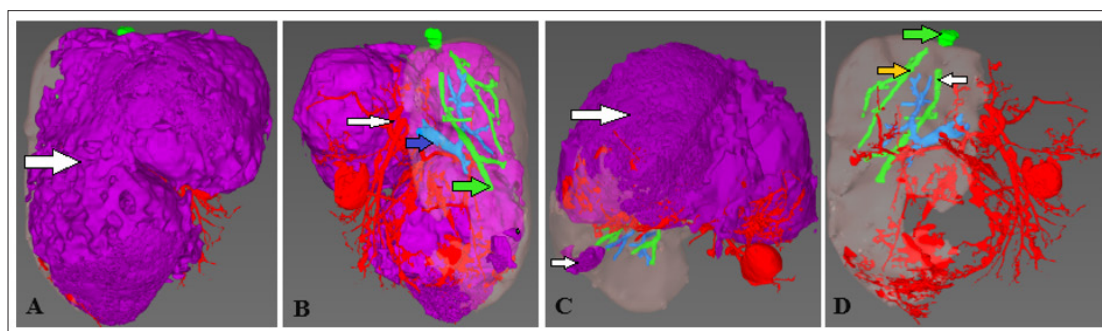


Figure 2b: 3D Simulation System of Platform Marian Software before Surgery

A (Anterior surface): Hemangioma (white arrow); B (Posterior surface): branches of proper hepatic artery (white arrow), right portal hepatic vein (blue arrow); Hepatic vein (green outlined arrow); C. Hemangioma (white arrow); D. Inferior Vena Cava (green arrow), right hepatic vein (yellow arrow); middle hepatic vein (white arrow).

Operation

Intraoperative investigation revealed the two large hepatic hemangiomas in the left lobe and the 5th-6th liver segment. The hepatic tumorectomy of the left liver and the 5th-6th liver segment was performed following the standard technique by using the clamp-crush technique for parenchymal transection. After completion of hepatectomy, an 8F feeding tube was inserted into the common bile duct for further (Figure 3). The left and right subphrenic drainage tubes were placed before abdominal wound closure to monitor the postoperative bile leakage and hemorrhage. The total blood loss was about 1000 ml with 700 ml of intraoperative red blood cell transfusion, and the duration of the operation was approximately 360 minutes. The cholangiography through biliary feeding tube drainage on day six after liver resection confirmed no evidence of bile leakage, and the abdominal CT scan with contrast on the same day showed a small amount of fluid in the right pleura and no signs of severe complications (Figure 4). The macroscopic examination of the resected specimens revealed two giant brownish lesions. Microscopic findings showed large spaces filled with blood inside and lined by endothelium. Histological examination confirmed the diagnosis of cavernous hepatic hemangioma (Figure 5). The patient was discharged on postoperative day 8.

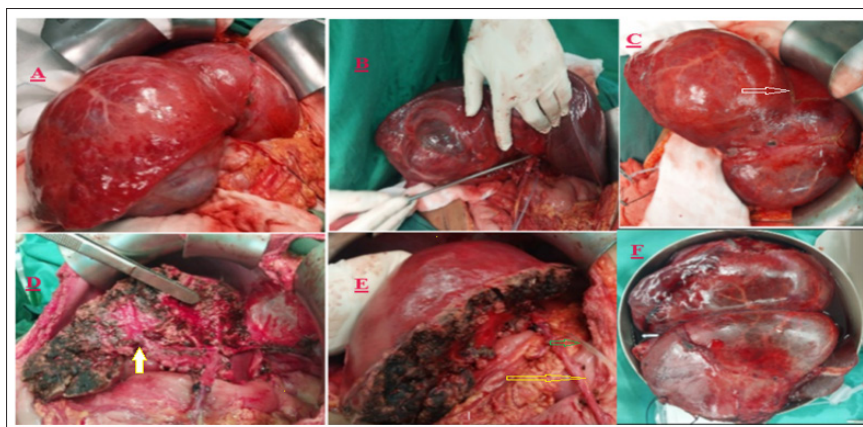


Figure 3: Intraoperative Images

A: Giant Hemangioma, B: Ligation and Cut of Small Pedicles from the Left Pedicle (Dissected from the Teres Ligament), C: Border of Tumor on Segment VIII (White Arrow), D: Pedicle of Segment VIII (Yellow outlined Arrow). E: The Teres Ligament (Yellow Outlined Arrow), Feeding Tube Inserted into the common Bile Duct (Green outlined arrow). F: Resected HH (two separate hemangiomas)

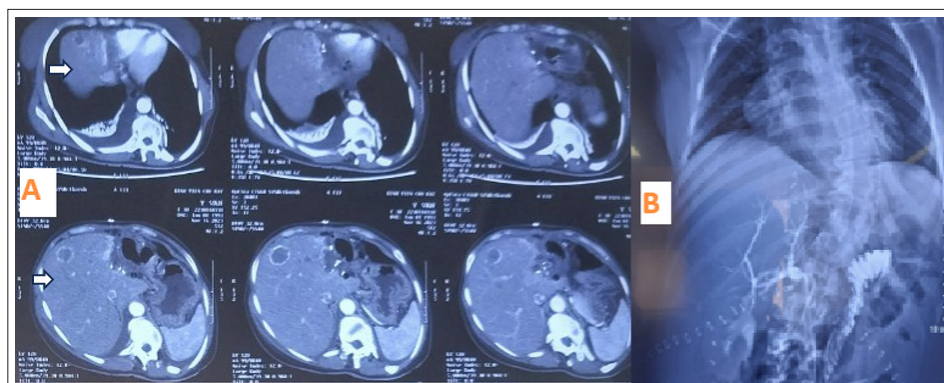


Figure 4: A: Postoperative Abdominal Ct scan with Contrast Showed Some Small Remaining Hepatic Hemangiomas (White Arrow) And B: Cholangiography (On Day Six Post-Operation) In Patient with Thoracic Scoliosis.

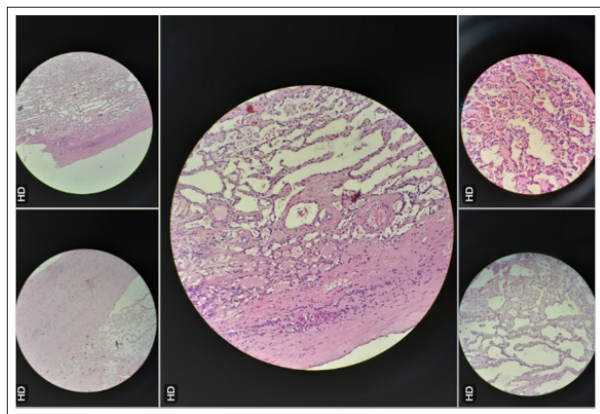


Figure 5: Microscopic Images Show Large Spaces Filled with Blood Inside and Lined by Endothelium

Discussion

HH is a benign tumor of the liver that is mostly incidentally diagnosed. Most people with HH show no signs or symptoms, and most hepatic hemangiomas are small, stable, and do not require treatment [6]. However, giant hemangiomas (>10cm) are symptomatic in over 80% of cases that affect a patient's life, owing to the compression of nearby organs, or can cause severe complications like intra-tumoral bleeding and spontaneous or iatrogenic rupture [7]. Therefore, treatments are offered to improve quality of life or prevent complications.

There are some treatments for HH, such as RFA, MWA, TAE, radiotherapy, and surgery. Among these treatments, surgical treatment is proven, considered more effective, and widely accepted. It can also be the first choice for most cases of giant symptomatic hepatic hemangiomas [4-8]. Previous studies also revealed that only 2% of overall HH cases are suitable for surgical treatment. However, symptomatic hepatic hemangiomas are rising by about 18-57% for this indication in the literature [7].

Due to recent developments in minimally invasive interventional techniques, some giant HHs can be treated by ablation or TAE effectively and gradually replaced as an alternative approach to surgical resection for treating HH when surgery is not feasible or is not accepted by the patient [4]. A multicenter retrospective analysis of 291 HH patients who underwent RFA revealed the effectiveness of this procedure. However, the study also showed some complications and recommended RFA for giant HH that measured less than 10 cm in diameter. However, this author also reported that three of the 291 cases of this study occurred in tumor rupture after undergoing RFA for HH, located on the surface of the liver [9]. Therefore, the site of HH is essential for treatment options.

HH is reported to be more common in female patients, and female sex hormones were involved in tumor growth in previous studies. In our case, a young pregnant woman, the rapid enlargement of the HH could occur during her pregnancy, and the HH was giant and protruded on the surface of the liver, it would be easy to cause bleeding if we underwent RFA. Otherwise, we assumed that these giant HH could be resectable based on the three-D simulation, which showed detailed hepatic vascular anatomy, its relation with the HH, and predicted the remaining liver volume to be enough for the patient if the two biggest hemangiomas were resected. Therefore, we decided to indicate liver resection for this patient.

Stanley was the first one to demonstrate TAE with polyvinyl alcohol to treat HH successfully in 1983 [10]. After that, many

studies reported the effectiveness and safety of this therapy in HH [11,12]. But, TAE is not yet a curative treatment according to Gao J because of recurrence owing to vascular recanalization⁸. Therefore, liver resection should be chosen for giant HH.

The first case of HH was performed liver resection performed in 1987 by Schwartz [12]. Then, many reports on surgical management in HH were presented. Koszka demonstrated a giant hemangioma measuring 40 cm in diameter and underwent left hepatectomy safely [13]. Sometimes, TAE was able to be used for the treatment of HH but not practical for giant HH; Makal described a case of HH in the left lobe 30 cm in size treated by surgical management after doing embolization unsuccessfully and noted that giant hepatic hemangioma should perform surgical treatment even if they do not have any symptoms and the size of tumor should also be considered in these patients [6]. Occasionally, TAE can be a good supportive therapy in giant HH to decrease blood loss before hepatectomy [14]. In addition, the effectiveness of TAE is also based on the type of blood supply of HH [4]. In another study of 373 patients with giant and enormous HH who underwent liver resection, Dong Z assumed that liver resection or enucleation could be performed safely for giant hemangioma. However, he suggested being careful with enormous HH because of a longer operation time and more intraoperative bleeding compared to giant HH [5-15].

Conclusion

Most HHs are small, asymptomatic, and not required to be treated. However, symptomatic giant HHs need to be treated to improve their quality of life and avoid the risk of serious complications. Treatment options should be weighted based on careful clinical and morphological features evaluation to do the best. Herein, we describe a young female patient with giant HH accompanying congenital thoracic scoliosis successfully treated with surgical resection. We suggest that patients who have symptomatic giant HH accompanying congenital thoracic scoliosis should undergo surgical treatment and three-dimensional simulation before surgery.

Conflict of Interest

The authors declare that they have no competing interests

Ethical Approval

This report was prepared in accordance with the ethical standards of the institutional ethics committee and with the 1964 Helsinki Declaration. Our hospital does not require ethical approval for reporting individual cases or case series.

Informed Consent

Written informed consent was obtained from the patient for publication of this case report.

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