



CASE REPORT / ПРИКАЗ БОЛЕСНИКА

Giant left lobe hemangioma of the liver misdiagnosed for splenomegaly

Borislav Tošković^{1,2}, Vladimir Milosavljević¹, Matija Buzejić³, Nataša Stanisavljević^{2,4}, Darko Zdravković^{2,5}

¹Bežanijska Kosa University Hospital Medical Center, Department for HPB Surgery, Belgrade, Serbia;

²University of Belgrade, Faculty of Medicine, Department for Surgery with Anesthesiology, Belgrade, Serbia;

³University Clinical Center of Serbia, Clinic for Endocrine Surgery, Belgrade, Serbia;

⁴Bežanijska Kosa University Hospital Medical Center, Department for Hematology, Belgrade, Serbia;

⁵Bežanijska Kosa University Hospital Medical Center, Department for Oncology Surgery, Belgrade, Serbia

SUMMARY

Introduction Most patients with liver hemangiomas are unrecognized, when symptoms occur it is usually due to the size of the hemangioma. Hemangioma of the liver are benign tumors which affects women more often. Surgical indications for liver resection remain unclear.

Case outline We present a patient with a giant hemangioma of the left liver lobe that was misdiagnosed in a primary care unit. The patient underwent resection of the left liver lobe and fully recovered after several days.

Conclusion Symptoms, size, and risk of rupture should be considered when decision for surgery is made. Linear stapler can be useful especially when left and middle hepatic vein have common trunk.

Keywords: liver; hemangioma; surgery; liver resection

INTRODUCTION

Liver hemangioma is one of the most common benign lesions of the liver, which can affect 20–30% of the general population. It is more diagnosed in women than in men with ratio up to 5:1 [1]. Hemangiomas are usually diagnosed incidentally on computer tomography (CT), magnetic resonance (MRI), or ultrasound (US) of the abdomen. They usually grow silently but it can be manifested as abdominal pain if it grows larger than 10 cm. Hemangiomas larger than 4 cm are diagnosed as giant liver hemangiomas based on literature [2, 3]. Pathogenesis is not clear, but it is a congenital vascular malformation or a hamartoma [4]. At histopathology exam, it is usually revealed as a mesenchymal lesion consisting of blood-filled vascular cavities of different sizes, surrounded by a simple layer of flat endothelial cells, supported by a fibrous connective tissue.

The aim of our work is to present a rare giant hemangioma of the liver initially interpreted as splenomegaly. We also present the operative technique and a review of the current literature.

CASE REPORT

A 45-year-old female patient was admitted to the hospital at the Hematology Department for further examination as massive splenomegaly was verified on an abdominal US. After

admission to our hospital, we performed a CT scan of the abdomen, which showed a discrepancy in relation to the US finding of the abdomen. Namely, a liver tumor in the left lobe was verified, after which an MRI with retrograde cholangiopancreatography was performed and the CT findings were confirmed – more precisely, a giant tumor of the left lobe of the liver was verified, occupying the left hypochondriac and left lumbar quadrants of the abdomen. The tumor around 22 cm in diameter dislocated the spleen towards the pelvis (Figure 1). The patient reported discomfort in the abdomen and decreased appetite. Laboratory values were between the reference ranges, including alpha fetoprotein, carcinoembryonic antigen, and cancer antigen 19-9. Based on the performed diagnostics and laboratory parameters, the conclusion was that it has been a giant hemangioma of the liver. An indication for operative treatment was established. Firstly, embolization of the left branch of the hepatic artery was attempted in order to possibly reduce the volume of the tumor, but, for technical reasons, the procedure was not performed successfully. Since the tumor was bigger than 20 cm in diameter, surgical team opted for “J” laparotomy. After incision, when we approached the abdomen, a giant-left-lobe liver was presented that occupied the left side of the abdomen. The next step was extrahepatic dissection, after identification of the left side of the hepatoduodenal ligament. The left hepatic artery was taped, followed by the left portal vein being taped, and dissected

Received • Примљено:

September 4, 2023

Accepted • Прихваћено:

October 20, 2023

Online first: November 10, 2023

Correspondence to:

Vladimir MILOSAVLJEVIĆ
Bežanijska Kosa University
Hospital Medical Center
Department for HPB Surgery
Dr Žorža Matea bb
11080 Belgrade
Serbia

milosavljevicvladimir10@gmail.com

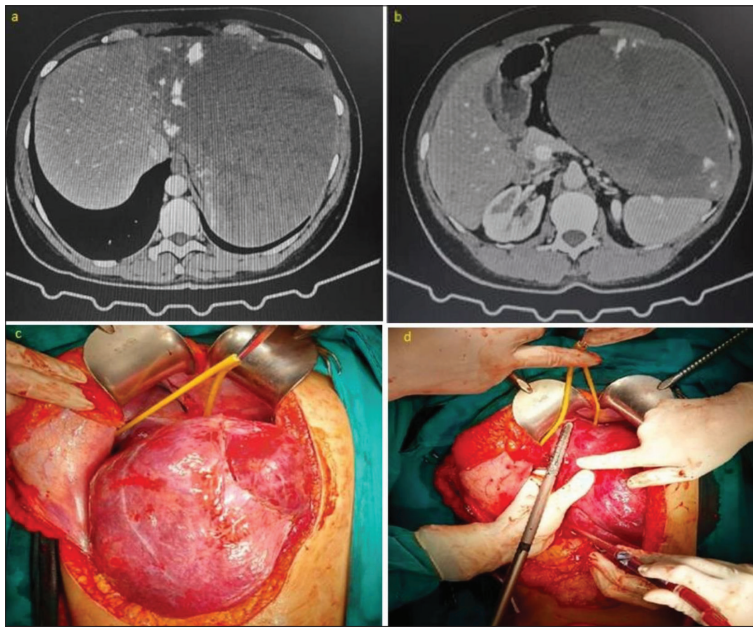


Figure 1. a, b – Magnetic resonance showing enlarged left lobe of the liver due to hemangioma; c, d – intraoperative finding and placed linear stapler

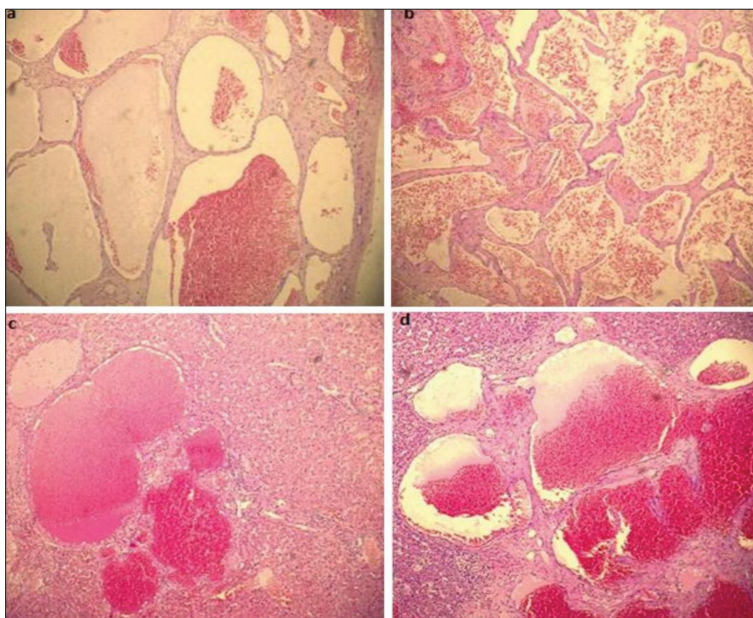


Figure 2. a, b, c, d – Pathological images, showing proliferation of blood vessels with thinned walls covered by endothelium without atypia (H&E, 5 ×)

as far as the root of the right portal vein. The left hepatic duct was identified just above the left portal vein. The falciform, right and left coronary, and left triangular ligaments were incised to mobilize the left lateral section. The inferior vena cava ligament, which fixes the caudate lobe to the cava from behind, was ligated and divided. After these steps, parenchymal dissection was begun on the inferior surface and was continued along the middle hepatic vein with linear stapler (Figure 1). The left and the middle hepatic vein had a common trunk, so the stapler was very useful. The tumor weight was 3200 g after being pulled out from the abdomen. The postoperative period was uneventful, and the patient was discharged from hospital on the fifth postoperative day. A histopathological finding revealed that the

tumor consisted mainly of a large number of abnormally dilated blood sinuses, there were no signs of endothelial atypia (Figure 2).

DISCUSSION

Liver hemangioma is the most common hepatic tumor, and it more often affects women than men. The reason for this sex-related occurrence inequality may be that some hemangiomas express estrogen receptors – those hemangiomas tend to grow during pregnancy and oral contraceptive drug use [1, 5]. Previously, hepatic hemangiomas larger than 5 cm in diameter were considered giant hepatic hemangiomas. However, data from the active literature indicate that hemangiomas with a diameter greater than 10 cm should be considered giant, which is more in line with the characteristics of the tumor and the requirements for diagnosis and treatment [6]. Extremely giant hepatic hemangiomas (> 10 cm) are rare and generally asymptomatic. When the symptomatology is present, it is mostly related to the compressive effect on the surrounding organovascular structures, resulting in a feeling of discomfort in the abdomen, pain, nausea, etc. [7, 8].

The patient we are presenting was of medium osteomuscular structure, and her chief problem was the size of the liver tumor. Namely, she had a feeling of heaviness and bloating in her stomach, inability to lie on her left side, frequent nausea.

The diagnostic methods for hepatic hemangioma include US, CT, MRI, scintigraphy, and positron-emission tomography combined with CT, angiography [9]. At US it usually presents as a hyperechogenic lesion with posterior acoustic enhancement. In CT, the density of the lesion is the same as the vessels. In MRI, it presents as homogenous and hyperintense on T2-weighted images, hypointense on T1-weighted images [10, 11].

In our case, we applied a CT protocol for hemangioma [12], and also performed MRI of the abdomen with retrograde cholangiopancreatography in order to fully assess the relationship of the tumor with the biliary ducts and surrounding organovascular structures.

In addition to abdominal discomfort and the size of the lesion, indications for surgery include spontaneous or traumatic rupture, rapidly enlarging lesions, Kasabach–Merritt syndrome, and unclear diagnosis (suspect of malignancy) [8, 13]. Treatments may be radiofrequency ablation, monoclonal antibody therapy, radiation therapy, trans-arterial embolization, interferon therapy, liver transplantation, and surgical procedures (enucleation or resection) [14, 15].

General consideration between surgical approaches is that enucleation is performed in a shorter operative time, but the capsule of hemangioma rupture bleeding is hard to get under control; in contrast, during resection when left hepatic vein is occluded, and pringle maneuver is made along with decreased central venous pressure, operative time can be shortened, and bleeding can be less. Also, intraoperative venous bleeding can be reduced by lowering central venous pressure and portal vein pressure by reducing collateral vessel filling, which helps to reduce intraoperative venous bleeding [16].

Since the hemangioma in our patient occupied almost the entire left lobe of the liver, along with the fact that the arterial embolization was not successfully performed, we decided on surgical treatment, i.e. resection of the liver using the stapler technique.

Hemangioma of the liver is a benign disease, which, however, can cause certain problems that impair the

quality of life of patients. Operative treatment or resection of the liver should be carefully considered when the tumor grows, if it is larger than 10 cm in diameter and if patients have pronounced symptoms. Liver transplantation should be considered in fewer cases if a giant hemangioma is present in both lobes (due to localization). Liver resection is a safe and effective surgical procedure. Also, the application of a linear stapler can be useful especially when left and middle hepatic vein have a common trunk.

Ethical standards: All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Conflict of interest: None declared.

REFERENCES

1. Trotter JF, Everson GT. Benign focal lesions of the liver. *Clin Liver Dis.* 2001;5(1):17–42, v. [DOI: 10.1016/s1089-3261(05)70152-5] [PMID: 11218914]
2. Xie QS, Chen ZX, Zhao YJ, Gu H, Geng XP, Liu FB. Outcomes of surgery for giant hepatic hemangioma. *BMC Surg.* 2021;21(1):186. [DOI: 10.1186/s12893-021-01185-4] [PMID: 33832476]
3. Sakamoto Y, Kokudo N, Watadani T, Shibahara J, Yamamoto M, Yamaue H; Japanese Society of Hepato-Biliary-Pancreatic Surgery. Proposal of size-based surgical indication criteria for liver hemangioma based on a nationwide survey in Japan. *J Hepatobiliary Pancreat Sci.* 2017;24(7):417–25. [DOI: 10.1002/jhbp.464] [PMID: 28516570]
4. Makal GB, Sonbahar BÇ, Özalp N. Surgical Treatment of Giant Liver Hemangioma, Case Report and Literature Review. *Sisli Etfal Hastan Tip Bul.* 2019;53(3):318–21. [DOI: 10.14744/SEMB.2017.09815] [PMID: 32377104]
5. Reddy KR, Kligerman S, Levi J, Livingstone A, Molina E, Franceschi D, et al. Benign and solid tumors of the liver: relationship to sex, age, size of tumors, and outcome. *Am Surg.* 2001;67(2):173–8. [PMID: 11243545]
6. Dong Z, Fang K, Sui C, Guo J, Dai B, Geng L, et al. The surgical outcomes and risk factors of giant hepatic haemangiomas: a single centre experience. *BMC Surg.* 2022;22(1):278. [DOI: 10.1186/s12893-022-01721-w] [PMID: 35843944]
7. Archer S, Ferreira AT, Rocha M, Pedrote I. Extremely giant liver hemangioma: a case beyond the norms. *Rev Esp Enferm Dig.* 2023 Aug 4. [DOI: 10.17235/reed.2023.9877/2023] Epub ahead of print. [PMID: 37539532]
8. Amsiguine N, Imrani K, El Houss S, Rguieg N, El Messaoudi I, Moatassim Billah N, et al. Kasabach-Merritt syndrome complicating a giant hemangioma of the liver: A case report. *Radiol Case Rep.* 2023;18(6):2183–5. [DOI: 10.1016/j.radcr.2023.01.084] [PMID: 37101891]
9. Zhao Y, Li XP, Hu YY, Jiang JC, Zhao LJ. Liver transplantation for giant hemangioma of the liver: A case report and review of the literature. *Front Med (Lausanne).* 2022;9:985181. [DOI: 10.3389/fmed.2022.985181] [PMID: 36186795]
10. Lim KJ, Kim KW, Jeong WK, Kim SY, Jang YJ, Yang S, et al. Colour Doppler sonography of hepatic haemangiomas with arterioportal shunts. *Br J Radiol.* 2012;85(1010):142–6. [DOI: 10.1259/bjr/96605786] [PMID: 21385916]
11. Jiang T, Zhao Z, Cai Z, Shen C, Zhang B. Case Report: Giant abdominal hemangioma originating from the liver. *Front Oncol.* 2023;13:1165195. [DOI: 10.3389/fonc.2023.1165195] [PMID: 37588097]
12. Mamone G, Di Piazza A, Carollo V, Cannataci C, Cortis K, Bartolotta TV, et al. Imaging of hepatic hemangioma: from A to Z. *Abdom Radiol (NY).* 2020;45(3):672–91. [DOI: 10.1007/s00261-019-02294-8] [PMID: 31686179]
13. Toro A, Mahfouz AE, Ardiri A, Malaguarnera M, Malaguarnera G, Loria F, et al. What is changing in indications and treatment of hepatic hemangiomas. A review. *Ann Hepatol.* 2014;13(4):327–39. [PMID: 24927603]
14. Meguro M, Soejima Y, Taketomi A, Ikegami T, Yamashita Y, Harada N, et al. Living donor liver transplantation in a patient with giant hepatic hemangioma complicated by Kasabach-Merritt syndrome: report of a case. *Surg Today.* 2008;38(5):463–8. [DOI: 10.1007/s00595-007-3623-4] [PMID: 18560973]
15. Obana A, Sato Y. Autologous Partial Liver Transplantation for a Symptomatic Giant Hepatic Hemangioma. A Case Report. *Transplant Proc.* 2022;54(10):2787–90. [DOI: 10.1016/j.transproceed.2022.09.025] [PMID: 36404155]
16. Ariizumi SI, Kotera Y, Yamashita S, Omori A, Kato T, Shibuya G, et al. Debulking of giant liver hemangiomas with severe symptoms: a case report. *Surg Case Rep.* 2020;6(1):195. [DOI: 10.1186/s40792-020-00960-4] [PMID: 32748049]

Велики хемангиом левог лобуса јетре иницијално протумачен као спленомегагија

Борислав Тошковић^{1,2}, Владимир Милосављевић¹, Матија Бузејић³, Наташа Станисављевић^{2,4}, Дарко Здравковић^{2,5}

¹Универзитетска болница „Бежанијска коса“, Одељење хепатобилијарне хирургије, Београд, Србија;

²Универзитет у Београду, Медицински факултет, Катедра за хирургију и анестезиологију, Београд, Србија;

³Универзитетски клинички центар Србије, Клиника за ендокрину хирургију, Београд, Србија;

⁴Универзитетска болница „Бежанијска коса“, Одељење хематологије, Београд, Србија;

⁵Универзитетска болница „Бежанијска коса“, Одељење онколошке хирургије, Београд, Србија

САЖЕТАК

Увод Већина хемангиома јетре је непрепозната код болесника, а када се симптоми појаве, то је углавном због величине промене. Хемангиоми јетре су бенигни тумори, који су чешћи у женској популацији. Консензус око одлуке о операцији и ресекцији јетре не постоји, изузев када су у питању руптура хематома и крварење.

Приказ болесника Болесница је примљена у болницу под сумњом на спленомегагију, а даљим радиолошким испитивањем доказан је велики тумор левог режња јетре.

С обзиром на величину промене, учињена је ресекција левог режња јетре са хемангиомом. Болесница је у добром општем стању отпуштена из болнице четвртог постоперативног дана.

Закључак Величина, симптоми и ризик од руптуре хемангиома треба да буду главни критеријуми када се доноси одлука о оперативном лечењу хемангиома јетре. Коришћење стаплера може бити корисно приликом ресекције, посебно када лева и средња хепатична вена имају заједнички трункус.

Кључне речи: јетра; хемангиом; хирургија; ресекција јетре