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Case Report / Приказ болесника

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Misdiagnosed giant left lobe hemangioma of liver for splenomegaly

Велики хемангиом левог лобуса јетре иницијално протумачен као спленомегалија

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SUMMARY

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

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

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

Keywords: liver; hemangioma; surgery; liver resection

Сажетак

Увод Болесница је примљена у болницу под сумњом спленомегалију, на 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 као 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 ресекција посебно када лева и средња хепатична вена имају заједнички трункус. Кључне речи: јетра; хемангиом; хирургија; ресекција јетре

INTRODUCTION

Liver hemangioma is one of the most common benign lesions of the liver, which can affect 20 to 30 % of general population. It is more diagnosed in women than in men with ratio up to 5:1 respectively [1]. Hemangiomas are usually diagnosed incidentally on computer tomography (CT), magnetic resonance (MRI) or ultrasound (US) of abdomen. They usually grow silently but it can be manifested as abdominal pain if it grows larger than 10 cm. Hemangioma larger than 4 cm are diagnosed as giant liver hemangioma based on literature [2, 3]. Pathogenesis is not clear, but it is congenital vascular malformation or hamartoma [4]. At histopathology exam usually it is revealed as mesenchymal lesion consisting of blood-filled vascular cavities of different size, 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 which was 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 in the hematology department for further examination because massive splenomegaly was verified on an abdominal ultrasound. After admission to our hospital, we performed a CT scan of the abdomen and , which showed a discrepancy in relation to the ultrasound finding of the abdomen. Namely, a liver tumor in the left lobe was verified, after which a magnetic resonance imaging with retrograde cholangiopancreatography (MRCP) 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. Tumor dislocated spleen towards pelvis, and it was around 22 cm id diameter (Image 1.). Patient reported discomfort in abdomen and decreased appetite. Laboratory values were between reference range so as alpha fetoprotein, carcinoembryonic antigen, and cancer antigen 19-9. Based on the performed diagnostics and laboratory parameters, the conclusion is that it is a giant hemangioma of the liver. An indication for operative treatment was established. First, 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 tumor was bigger than 20 cm in diameter, surgical team decided for J laparotomy. After incision, when we approached abdomen giant left lobe liver was presented that occupied left side of abdomen. 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 as far as the root of the right portal vein. The left hepatic duct (LHD) 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 those steps parenchymal dissection was begun on the inferior surface and was continued along the middle hepatic vein with linear stapler (Figure 1). Left and the middle hepatic vain had common trunk so stapler was very useful. Tumor weight was 3200 gr after being pulled out from abdomen. Postoperative period was uneventful, and the patient was discharged from hospital at the 5th postoperative day. Histopathological finding revealed that 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 often affects women than men. Reason for this gender occurrence inequality may be that some hemangiomas express estrogen receptors- those hemangiomas tend to grow during pregnancy and oral contraceptive drug usage [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 and then there is a feeling of discomfort in the abdomen, pain, nausea, and so on [7, 8].

The patient we are presenting was of medium ostemuscular structure, so her biggest 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 ultrasonography, CT, MRI, scintigraphy, and positron-emission tomography combined with CT, angiography [9]. At US it usually presented as 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 MR of the abdomen with retrograde cholangiopancreatography in order to fully assess the relationship of the tumor with the biliary ducts and surrounding organovascular structures.

Besides abdominal discomfort and 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 (RFA), monoclonal antibody therapy, radiation therapy (RT), trans-arterial embolization (TAE), 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 capsule of hemangioma rupture it is hard to get bleeding under control; on contrary 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, due to all of the above, 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 common trunk.

Ethical standards: All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or 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;5:17–42. 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 Apr 8;21(1):186. doi: 10.1186/s12893-021-01185-4. PMID: 33832476
- Sakamoto Y, Kokudo N, Watadani T, Shibahara J, Yamamoto M, Yamaue H, et al. Proposal of size-based surgical indication criteria for liver hemangioma based on a nationwide survey in Japan. J Hepatobiliary pancreat Sci. 2017;24:417–25 doi: 10.1002/jhbp.464. Epub 2017 Jun 22. PMID: 28516570.
- Makal GB, Sonbahar BÇ, Özalp N. Surgical Treatment of Giant Liver Hemangioma, Case Report and Literature Review. Sisli Etfal Hastan Tip Bul. 2019 Aug 28;53(3):318-321. doi: 10.14744/SEMB.2017.09815. PMID: 32377104; PMCID: PMC7192281.
- 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 Feb;67(2):173-8. PMID: 11243545...
- 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 Jul 17;22(1):278. doi: 10.1186/s12893-022-01721w. PMID: 35843944
- 7. Archer S, Ferreira AT, Rocha M, Pedroto 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.
- 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 Apr 11;18(6):2183-2185. doi: 10.1016/j.radcr.2023.01.084. PMID: 37101891
- Zhao Y, Li X-P, Hu Y-Y, Jiang J-C and Zhao L-J. Liver transplantation for giant hemangioma of the liver: A case report and review of the literature. Front. Med. 2022. 9:985181. doi: 10.3389/fmed.2022.985181. PMID: 36186795; PMCID: PMC9523786.
- 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) 85:142–6. doi: 10.1259/bjr/96605786. Epub 2011 Mar 8. PMID: 21385916; PMCID: PMC3473947.
- 11. Jiang T, Zhao Z, Cai Z, Shen C, Zhang B. Case Report: Giant abdominal hemangioma originating from the liver. Front Oncol. 2023 Jul 31;13:1165195. doi: 10.3389/fonc.2023.1165195. PMID: 37588097; PMCID.
- 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 Mar;45(3):672-691. 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) 13:327–39. 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) 38:463–8. doi: 10.1007/s00595-007-3623-4. Epub 2008 Apr 30. PMID: 18560973.
- Obana A, Sato Y. Autologous Partial Liver Transplantation for a Symptomatic Giant Hepatic Hemangioma. A Case Report. Transplant Proc. 2022 Dec;54(10):2787-2790. doi: 10.1016/j.transproceed.2022.09.025. Epub 2022 Nov 18. PMID: 36404155.
- 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 Aug 3;6(1):195. doi: 10.1186/s40792-020-00960-4. PMID: 32748049



Figure 1. a, b – Magnetic resonance showing enlarged left lobe of 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, $5 \times$)