Contents lists available at ScienceDirect



International Journal of Surgery Case Reports





Case report Combined surgical treatment of giant cavernous hepatic hemangioma: A case report

R.A. Goncharuk, Zh.A. Rakhmonov^{*}, K.V. Stegnii, A.A. Krekoten, I.V. Shulga, E.R. Dvoinikova

Medical Center of the Far Eastern Federal University, Aiax Settlement, 10, FEFU Campus, Building 25, Vladivostok, Russian Island 690922, Russian Federation

ARTICLE INFO	A B S T R A C T
<i>Keywords</i> : Hepatic hemangioma Transarterial embolization Hemihepatectomy Hepatic resection Case report	Introduction and importance: Hepatic hemangiomas are the most common benign liver tumors, which in most cases are small in size and do not require specific treatment. Giant hepatic hemangiomas are a rare pathology. The purpose of this report is to demonstrate the case of combined treatment of giant hepatic hemangioma. <i>Case presentation:</i> A man of the Caucasian race, 37 years old, appealed to the clinic with a complaint about the presence of palpable abdominal mass. During the examination a giant cavernous hemangioma of the left lobe of the liver with a vertical size of up to 210 mm was discovered. Endovascular transarterial embolization (ETE) of tumor-feeding hepatic artery followed by radical surgery on mass removal is performed. <i>Clinical discussion:</i> Giant hemangiomas often cause compression of nearby structures and cause symptoms such as pain, abdominal discomfort, nausea, early food saturation, etc. Cases of spontaneous rupture of cavernous hemangioma, including fatal outcome, are also described. We suggest that some patients should go through surgical treatment even if they do not have any complaint. <i>Conclusions:</i> Our experience shows that endovascular embolization of the hepatic arteries can be effectively applied as a pre-surgical preparation method in order to reduce hemangioma size and blood loss volume during surgical intervention.

1. Introduction

Hepatic hemangiomas are the most common benign liver tumors. The frequency of occurrence in the population reaches from 0.4 to 20% [1,2]. One of the most current issues is the diagnosis of giant hepatic hemangiomas. Although, imaging characteristics of hemangiomas at computed tomography (CT) and magnetic resonance imaging (MRI) are well known: progressive peripheral nodal enhancement at dynamic cross-sectional studies and delayed centripetal filling at intravenous contrast examination, there are a wide range of atypical variants: hemangioma with centrifugal enhancement, hemangioma with calcificates, sclerosing hemangioma, etc., which cause difficulties in the diagnosis process [3,4]. For this reason, a possibility of biopsy of mass is considered. However, this method involves a high risk of hemorrhagic complications, as a result of which it is not recommended [2,5]. Cases of fatality after performing thin-needle aspiration biopsy of hepatic hemangioma are described [6].

Cases of spontaneous rupture of cavernous hemangioma [7], including fatal outcome [8], are also described. Moreover, in a study by

Yedibela et al. with the separation of patients into surgical treatment and dynamic follow-up groups, two cases with fatality were recorded in the follow-up group as a result of spontaneous hemangioma rupture [9].

The risk of spontaneous and traumatic hemangioma rupture is small. In case of progression of this complication, the probability of fatality is very high and is 70% [7,10].

Traditional approaches to treating hemangiomas are hepatic resection and mass enucleation. Transarterial embolization is currently an alternative option in choosing a method of surgical treatment for hepatic hemangiomas with a size of more than 15 cm [11–13].

We demonstrate a case of successful combined treatment of asymptomatic giant hepatic hemangioma, during which ETE of the tumorfeeding hepatic artery, followed by radical surgery on mass removal, was sequentially performed.

This case report has been reported in line with the SCARE 2020 criteria [14].

https://doi.org/10.1016/j.ijscr.2022.107012

Received 9 March 2022; Received in revised form 27 March 2022; Accepted 31 March 2022 Available online 2 April 2022

^{*} Corresponding author at: Aiax Settlement, 10, FEFU Campus, Building 10, Vladivostok, Russian Island 690922, Russian Federation. *E-mail address:* zhavokhirrakhmanov@gmail.com (Zh.A. Rakhmonov).

^{2210-2612/© 2022} The Authors. Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

2. Case report

A man of the Caucasian race, 37 years old, appealed to the clinic with a complaint about the presence of palpable abdominal mass. Ultrasound scanning detected large-sized mass ($21.0 \times 20.1 \text{ cm}$) from the left lobe of the liver. Notably, there were no symptoms and signs, the mass did not affect the patient's quality of life. Hereditary, social history without abnormalities.

On palpation of the abdominal cavity, a rounded, pain-free, immobile mass from the epigastric to the paraumbilical area is determined. Laboratory blood tests revealed an increase in total bilirubin levels to 1.67 (norm is <1.20 mg/dl) and fibrinogen to 735 (norm is 200-400 mg/dl). During CT examination of abdominal organs, a giant hypervascular mass was detected in the left lobe of the liver (S3, S4) 205 * 154 * 210 mm with the parenchyma pushed upwards, with close adherence to the stomach, duodenum, pancreas gland and gallbladder with signs of their pronounced compression. Compression of the stomach outside is confirmed by esophagogastroduodenoscopy (EGDS). The mass had an irregular shape, clear bulging contours with the presence of partitions and the formation of multiple cavities. The left lobular hepatic artery and its segmental branch of segment III are highly hypertrophied; the diameter of the left hepatic artery is up to 5–5.5 mm. The left portal vein is compressed and occluded by a tumor. The portal vein is expanded to 22 mm. At intravenous dynamic contrasting, there is an accumulation of contrast starting from the peripheral segments in the arterial phase with diffusion to the mass center in the delayed phase (Fig. 1). At MRI, a similar image with hypointense mass in the T1 mode, hyperintense mass in the T2 mode was obtained. The findings indicate a giant cavernous hemangioma.

Given the giant tumor size, upper gastrointestinal (GIT) organ compression, a possibility of a spontaneous or traumatic rupture of a hemangioma, followed by intra-abdominal bleeding, the benign nature of a disease and also complexity of ensuring access to hepatoduodenal ligament elements, it was decided at the first stage to perform the ETE of the hemangioma-feeding artery as pre-surgery preparation for reduction in volume of intraoperative blood loss and also reduction in size of mass and so that to ensure safety of future radical surgery.

Selective embolization of left hepatic artery branches is performed by microspheres "Embosphere 900-1200" and "Axium" spirals. The post-surgery period was without complications.

On the 24th day after ETE, a control CT of abdominal organs was performed in order to assess the dynamics of the mass sizes. The maximum vertical hemangioma dimension decreased to 150 mm (Fig. 2).

A significant decrease in tumor volume provided safe access to hepatoduodenal ligament elements, hepatic veins; on the 25th day after ETE, the mass was completely removed by performing left-sided hepatectomy through access of the "Mercedes" type, which is associated with the giant size of the hemangioma, as well as for the convenience and safety of working with large vascular bundles (Fig. 3). The intervention was completed by installing a hemostatic sponge on the liver stump and draining the abdominal cavity with two drainage tubes under the liver on the right. The duration of surgery was 225 min, intraoperative blood loss -200 ml. Morphological examination of removed medication confirmed the cavernous hepatic hemangioma. The post-surgery period was without complications, drainage was removed from abdomen on the 10th day, which was due to the need of control the bile leakage and prevention the biloma development. The patient in satisfactory condition was discharged on the 12th day after the surgery. During a control examination in 5 months, there are no complaints, control CT of abdominal organs without abnormalities.

3. Discussion

Cavernous hepatic hemangiomas are benign masses, which in most cases are small in size (<4 cm). As for the term "giant hemangioma", there is no generally accepted definition in the literature. According to some authors, tumors more than 4–5 cm in diameter [15,16] are considered giant, according to others – more than 10 cm [1]. Hemangiomas of small sizes are usually found by chance, have asymptomatic course and do not require treatment, only dynamic follow-up is enough [9]. Giant hemangiomas often cause compression of nearby structures and cause symptoms such as pain, abdominal discomfort, nausea, early food saturation, etc. [4].

The choice of method of surgery intervention is important. Several studies comparing mass enucleation with hepatic resection have demonstrated that enucleation is associated with lower mortality, shorter duration of surgery, less blood loss and fewer complications [17,18]. During enucleation, more hepatic parenchyma is saved [18]. However, a single-center study, which included 86 patients with hemangioma > 10 cm, conducted by Zhang et al., did not reveal a significant difference between enucleation and resection relative to the duration of surgery, amount of blood loss, complications and period of hospitalization [19].

Transarterial embolization of the tumor-feeding artery is currently an important option in the treatment of hemangiomas. In multicenter study, including 836 patients, conducted by Li Y. et al., the role of transarterial chemoembolization using pingyangmycin-lipiodol as a safe and effective method that significantly reduces the volume of hepatic hemangioma is proved [13]. According to other authors, ETE should not be used as an independent method of treating hepatic hemangioma, since it does not exclude possible complications and the probability of revascularization and relapse [2,12]. However, the publications note the important role of ETE as a pre-surgery preparation method to reduce intraoperative blood loss and tumor size.

The time interval between the performance of ETE and hepatic resection is the subject of debate. A number of authors propose to conduct interval CT monitoring to determine the size of hemangioma before considering the scope of radical intervention [11,20]. However, Zhou et al., and Seo H. et al. noted that increased interval between ETE



Fig. 1. Axial (A), sagittal (B), and coronal (C) images in CT.



Fig. 2. Axial (A), sagittal (B), and coronal (C) images in CT after ETE.

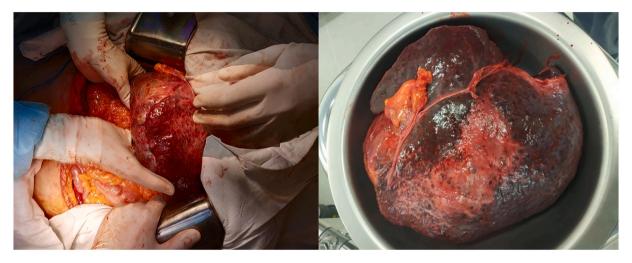


Fig. 3. Intraoperative and postoperative photographs of the tumor.

and hepatic resection leads to development of post-embolization pain syndrome, migration of embolization agent and development of intestinal obstruction [12,20]. The authors recommend the urgent surgery the following day after ETE.

In the present case, in connection with emergence of symptoms of an acute respiratory viral infection at the patient and an intense epidemiological situation on new coronavirus infection of SARS-CoV-2, surgery on mass removal was performed on the 25th day after ETE. Despite this, the above-described complications were not observed. Pre-surgery ETE contributed to a reduction in the size and volume of intraoperative blood loss (200 ml).

4. Conclusion

Our experience shows that endovascular embolization of the hepatic arteries can be effectively used as a method of pre-surgery preparation in order to reduce intraoperative blood loss and the size of giant hepatic hemangiomas.

For now an issue concerning the interval between the performance of ETE and hepatic resection remains unresolved. Further research is needed to clarify this situation.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Ethical approval

Ethical approval was not required.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Guarantor

Zh.A. Rakhmonov.

Research registration number

None.

CRediT authorship contribution statement

R.A. Goncharuk: diagnostics and treatment of the disease, the conception and design of the study, data interpretation. **Zh.A. Rakhmonov**: treatment of the disease, the conception and design of the study, data collection and analysis, writing the paper. **K.V. Stegnii**: data collection and analysis. **A.A. Krekoten**: data collection. **I.V. Shulga**:

data collection. **E.R. Dvoinikova**: data collection. All authors read, revised, and approved the final manuscript.

Declaration of competing interest

The author states that there is no conflict of interests.

References

- [1] M. Colombo, A. Forner, J. Ijzermans, V. Paradis, H. Reeves, V. Vilgrain, J. Zucman-Rossi, EASL clinical practice guidelines on the management of benign liver tumours, J. Hepatol. 65 (2016) 386–398, https://doi.org/10.1016/j. jhep.2016.04.001.
- [2] L.T. Hoekstra, M. Bieze, D. Erdogan, J.J.T.H. Roelofs, U.H.W. Beuers, T.M.V. Gulik, Management of giant liver hemangiomas: an update, Expert Rev. Gastroenterol. Hepatol. 7 (2013) 263–268, https://doi.org/10.1586/egh.13.10.
- [3] G. Mamone, A. Di Piazza, V. Carollo, C. Cannataci, K. Cortis, T.V. Bartolotta, R. Miraglia, Imaging of hepatic hemangioma: from A to Z, Abdom. Radiol. 45 (2020) 672–691, https://doi.org/10.1007/s00261-019-02294-8.
- K.J. Oldhafer, V. Habbel, K. Horling, G. Makridis, K.C. Wagner, Benign liver tumors, Visc. Med. 36 (2020) 292–303, https://doi.org/10.1159/000509145.
 M. Leon, L. Chavez, S. Surani, Hepatic hemangioma: what internists need to know,
- World J. Gastroenterol. 26 (2020) 11–20, https://doi.org/10.3748/wjg.v26.i11.11
 B.A. Terriff, R.G. Gibney, C.H. Scudamore, Fatality from fine-needle aspiration
- biops of a hepatic hemangioma, AJRAm J. Roentgenol. 154 (1990) 203–204, https://doi.org/10.2214/ajr.154.1.2104717.
- [7] W. Zhao, X. Guo, J. Dong, Spontaneous rupture of hepatic hemangioma: a case report and literature review, Int. J. Clin. Exp. Pathol. 8 (2015) 13426–13428.
- [8] M. Bel Hadj, M. Marzougui, N. Ben Abdeljelil, R. Dhouieb, A. Zakhama, A. Chadly, Spontaneous rupture of a hepatic cavernous hemangioma: a rare case of sudden unexpected death, Am. J. Forensic Med. Pathol. 41 (2020) 138–140, https://doi. org/10.1097/PAF.00000000000552.
- [9] S. Yedibela, S. Alibek, V. Müller, Ü. Aydin, M. Langheinrich, C. Lohmüller, W. Hohenberger, A. Perrakis, Management of hemangioma of the liver: surgical therapy or observation? World J. Surg. 37 (2013) 1303–1312, https://doi.org/ 10.1007/s00268-013-1904-1.
- [10] G. Bora, Surgical treatment of giant liver hemangioma; case report and literature review, SiSli Etfal Hastan, Tip Bul./Med. Bull. Sisli Hosp. 53 (2018) 318–321, https://doi.org/10.14744/semb.2017.09815.
- [11] N. Akamatsu, Y. Sugawara, M. Komagome, T. Ishida, N. Shin, N. Cho, F. Ozawa, D. Hashimoto, Giant liver hemangioma resected by trisectorectomy after efficient

volume reduction by transcatheter arterial embolization: a case report, J. Med. Case Rep. 4 (2010) 2–6, https://doi.org/10.1186/1752-1947-4-283.

- [12] H. Il Seo, H.J. Jo, M.S. Sim, S. Kim, Right trisegmentectomy with thoracoabdominal approach after transarterial embolization for giant hepatic hemangioma, World J. Gastroenterol. 15 (2009) 3437–3439, https://doi.org/ 10.3748/wjg.15.3437.
- [13] Y. Li, Y. Jia, S. Li, W. Wang, Z. Wang, Y. Wang, B. Liu, W. Wang, H. Chang, Z. Li, Transarterial chemoembolization of giant liver haemangioma: a multi-center study with 836 cases, Cell Biochem. Biophys. 73 (2015) 469–472, https://doi.org/ 10.1007/s12013-015-0680-y.
- [14] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, A. Kerwan, A. Thoma, A.J. Beamish, A. Noureldin, A. Rao, B. Vasudevan, B. Challacombe, B. Perakath, B. Kirshtein, B. Ekser, C.S. Pramesh, D.M. Laskin, D. Machado-Aranda, D. Miguel, D. Pagano, F. H. Millham, G. Roy, H. Kadioglu, I.J. Nixon, I. Mukherjee, J.A. McCaul, J.Chi-Yong Ngu, J. Albrecht, J.G. Rivas, K. Raveendran, L. Derbyshire, M.H. Ather, M. A. Thorat, M. Valmasoni, M. Bashashati, M. Chalkoo, N.Z. Teo, N. Raison, O. J. Muensterer, P.J. Bradley, P. Goel, P.S. Pai, R.Y. Afifi, R.D. Rosin, R. Coppola, R. Klappenbach, R. Wynn, R.L.De Wilde, S. Surani, S. Giordano, S. Massarut, S. G. Raja, S. Basu, S.A. Enam, T.G. Manning, T. Cross, V.K.L. Karanth, V. Kasivisvanathan, Z. Mei, The SCARE 2020 guideline: updating consensus Surgical CAse REport (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230, https:// doi.org/10.1016/j.ijsu.2020.10.034.
- [15] A. Amico, L. Mammino, S. Palmucci, R. Latino, P. Milone, G. Li Destri, B. Antonio, A. Di Cataldo, Giant hepatic hemangioma case report: when is it time for surgery? Ann. Med. Surg. 58 (2020) 4–7, https://doi.org/10.1016/j.amsu.2020.08.003.
- [16] K. Hanazaki, S. Kajikawa, A. Matsushita, T. Monma, M. Hiraguri, N. Koide, Y. Nimura, W. Adachi, J. Amano, Giant cavernous hemangioma of the liver: is tumor size a risk factor for hepatectomy? J. Hepato-Biliary-Pancreat. Surg. 6 (1999) 410–413, https://doi.org/10.1007/s005340050141.
- [17] W.L. Cheng, Y.Q. Qi, B. Wang, L. Tian, W. Huang, Y. Chen, Enucleation versus hepatectomy for giant hepatic haemangiomas: a meta-analysis, Ann. R. Coll. Surg. Engl. 99 (2017) 237–241, https://doi.org/10.1308/rcsann.2016.0349.
- [18] Y. Liu, X. Wei, K. Wang, Q. Shan, H. Dai, H. Xie, L. Zhou, X. Xu, S. Zheng, Enucleation versus anatomic resection for giant hepatic hemangioma: a metaanalysis, Gastrointest. Tumors 3 (2017) 153–162, https://doi.org/10.1159/ 000455846.
- [19] W. Zhang, Z.-Y. Huang, C.-S. Ke, C. Wu, Z.-W. Zhang, B.-X. Zhang, Y.-F. Chen, W.-G. Zhang, P. Zhu, X.-P. Chen, Surgical treatment of giant liver hemangioma larger than 10 cm: a single center's experience with 86 patients, Medicine (Baltimore) 94 (2015), e1420, https://doi.org/10.1097/MD.000000000001420.
- [20] J.X. Zhou, J.W. Huang, H. Wu, Y. Zeng, Successful liver resection in a giant hemangioma with intestinal obstruction after embolization, World J. Gastroenterol. 19 (2013) 2974–2978, https://doi.org/10.3748/wjg.v19.i19.2974.