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Successful treatment of a therapy-resistant, giant liver hemangioma with repeated bleomycin TACE: A case report

IMAGING

GABOR MOHOS^{1,2} , ADAM ZOLTAN FARKAS² ,
ANDRAS BIBOK² , DAVID ADAM KORDA² ,
OSZKAR HAHN³ , ATTILA DOROS² ,
PAL AKOS DEAK²  and DENES BALAZS HORVATHY^{2*} 

CASE REPORT



¹ Department of Radiology, Medical Imaging Centre, Semmelweis University 1082 Budapest, Korányi Sándor street 2., Hungary

² Department of Interventional Radiology, Heart and Vascular Centre, Semmelweis University 1122 Budapest, Határőr street 18., Hungary

³ Department of Surgery, Transplantation and Gastroenterology, Semmelweis University, 1082 Budapest, Üllői út 78., Hungary

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ABSTRACT

We report a case of a female patient in her 40s who presented with right upper quadrant pain and abdominal discomfort. Surgical removal was not possible due to the location of the lesion. Between 2021 and 2022, the patient had four additional sessions of conventional transarterial embolization (TAE) with Lipiodol, but none succeeded in sufficiently reducing the hemangioma's size or permanently improving the symptoms. In November 2022, we used transarterial chemoembolization (TACE) with a Bleomycin-Lipiodol emulsion. Follow-up MRI scans showed a gradual reduction in the hemangioma volume, but the symptoms only improved partially. Hence, the patient underwent a second bleomycin TACE embolization in November 2023. By August 2024, control showed a total volume reduction of 409 cm³ (56%) and significant symptom improvement. This case demonstrates that Bleomycin-Lipiodol TACE can effectively reduce the size of giant liver hemangiomas, even after the failure of other interventional therapies.

KEYWORDS

giant liver hemangioma, TACE, bleomycin

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*Corresponding author. Department of Interventional Radiology, Heart and Vascular Centre, Semmelweis University H-1122 Budapest, Határőr street 18., Hungary.
Tel.: +36 20 825 8591.
E-mail: horvathy.denes@semmelweis.hu

Introduction

Giant liver hemangiomas are venous vascular malformations, variably defined as lesions larger than 4–6 cm [1]. While often asymptomatic, they can cause abdominal pain in symptomatic cases. Management of symptomatic hemangiomas is controversial, with no established consensus [2–4]. In addition to surgical treatment, interventional options such as embolization and percutaneous sclerotization with a bleomycin-lipiodol emulsion are widely recognized for their efficacy [2, 5]. We report a case where a symptomatic giant liver hemangioma refractory to conventional TAE was successfully treated with transarterial bleomycin TACE.

Case presentation

Initially, In 2011, a 30-year-old female patient presented with right lower quadrant pain. Sonography incidentally revealed three small hemangiomas in her liver in segments five and

six. The patient was advised to follow up, but she did not return until 2015 when she experienced abdominal discomfort. Imaging revealed significant growth of the hemangiomas, with the largest in the right lobe measuring $79 \times 64 \times 61$ mm (251 cm³). Based on the symptoms, we assessed treatment options for the lesion adjacent to the right portal vein. The surgical opinion indicated that the only way to remove the giant hemangioma was through right hemihepatectomy, which was deemed too invasive for a benign lesion. For this reason, we decided to pursue interventional treatment for the giant hemangioma.

In June 2015, the patient underwent a superselective Transarterial Embolization (TAE) with 8 mL of Lipiodol, followed by a microwave ablation in December 2015 (multiple positions on 60 W, during 5–5 min). Initial size reduction was observed, but despite these interventions, follow-up imaging in 2016 showed growth of the lesion to 89 mm (188 cm³). The patient dropped out of follow-up, probably due to the lack of considerable symptoms. She presented again after a five-years, with recurring, sometimes severe abdominal pain in the right upper quadrant of the abdomen. By this time the lesion has grown to 673 cm³. At the beginning we believed that controlling the size (preventing further growth) could be a satisfactory outcome. For this reason, four TAE sessions have been performed between 2021 and 2022 using 10 mL of Lipiodol in each session. We observed a transient reduction in complaints following the treatments, which we considered a partial clinical success. However, no size reduction was achieved; on contrary the lesion showed further growth.

Due to persistent symptoms, in November 2022, we performed TACE with bleomycin-lipiodol emulsion (right brachial approach, 4F introducer, 4F H1 catheter and 2.7 F microcatheter). The preparation involved 15,000 unit of

bleomycin in 5 cm³ of saline mixed with 10 mL lipiodol. The procedure was performed via right brachial access and involved embolizing three feeding branches with a total of 12 mL of the emulsion. Subsequent MRIs in 2023 demonstrated a gradual volume reduction of 221 cm³ (28%), but the desired outcome was not fully achieved. In November 2023, we repeated the TACE using the same method, resulting in a total volume reduction of 409 cm³ (56%) by August 2024, over a 21-month follow-up period (Figs 1 and 2).

Discussion

Liver hemangiomas are often asymptomatic but can cause significant discomfort due to mass effect. Complications, such as rupture, are rare; therefore, without symptoms, observation is a safe choice to monitor if they grow. In symptomatic cases, on the other hand, there is currently no consensus on the optimal management. Surgery might seem too invasive, while conventional TAE might not decrease the size and symptoms sufficiently. Several studies have shown that Bleomycin-lipiodol emulsion, including hundreds of patients, is a safe and sufficient option for treating giant symptomatic cases [2, 6, 7].

In our present case, another interventional option, namely microwave thermoablation combined with conventional TAE, was used as the first treatment in 2015. This resulted in a significant size reduction (before treatment: 251 cm³, after treatment: 188 cm³), while the patient's original uncertain complaints also improved. Probably, this is the reason why she did not show up for follow-up until her complaints became unbearable in 2021 with a visible bulging of the tumor. By that time, the hemangioma was

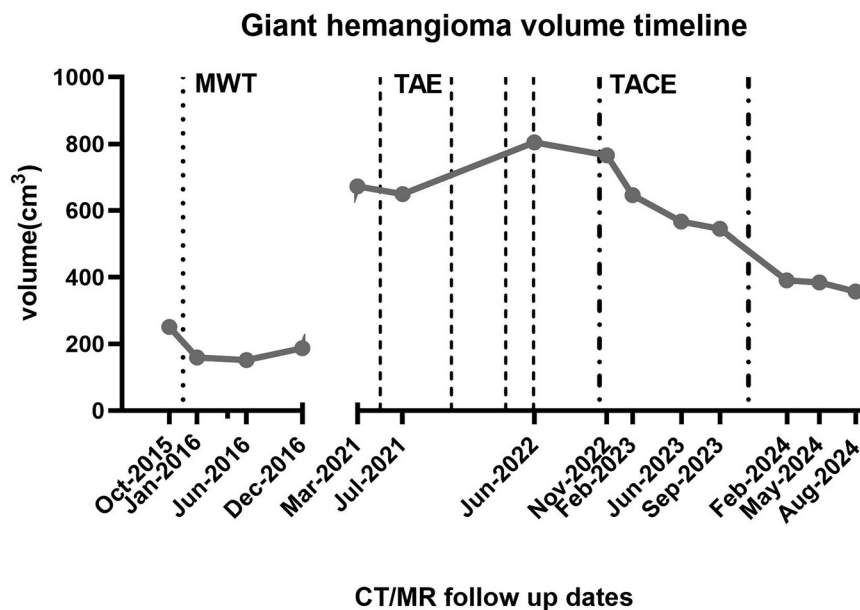


Fig. 1. Timeline of the giant hemangioma volume measurements. MWT indicates the time of the microwave thermoablation, dashed line indicates the TAE treatments, dash-dotted line indicates TACE treatments with bleomycin



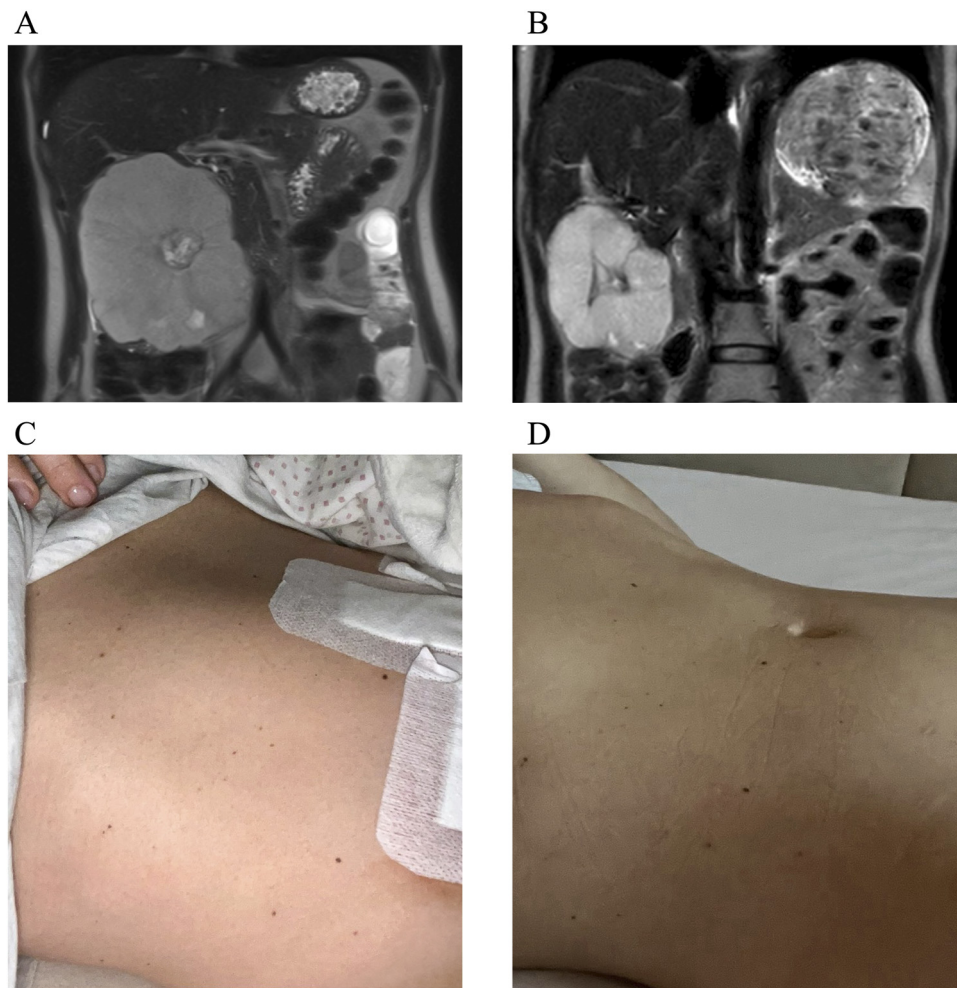


Fig. 2. Panel A shows coronal T2 MRI image of the giant haemangioma prior to the TACE treatments. Panel B shows coronal T2 MRI image 6 months after the second TACE. Panel C shows bulging of the haemangioma in supine position, while panel D demonstrates the cosmetic results six months after the last treatment

673 cm³. The patient reported that her symptoms are mild in the morning and worsen throughout the day.

The patient had been taking contraceptives prior to her illness; however, she discontinued them as soon as the growth of the hemangioma was confirmed in 2015. Based on this, we assume that contraceptive use played a minimal or negligible role in the growth of the hemangioma. It is more important to note that the hemangioma was located in segment 5 and it was exophytic, so there was no liver parenchyma caudal to the lesion. Additionally, the patient had a standing job, which could explain the enormous growth of the lesion and the complaints getting worse throughout the day. At this point, we have applied four sessions of conventional TAE [8], but the hemangioma kept growing, along with the symptoms. We chose conventional TAE because bleomycin TACE was less widespread back then, and we tried to avoid using a cytotoxic agent in a young patient with a benign lesion. While NBCA or particles could also have been considered

viable options, we determined that these materials would likely have remained permanently within the haemangioma, potentially stabilizing its size without achieving significant reduction. Given the lesion's pronounced bulging into the abdomen and the exophytic nature causing severe symptoms, achieving size reduction was considered essential.

Following bleomycin TACE, significant volume reduction was achieved, along with improved symptoms and cosmetic outcomes (Figs 1 and 2). After 2 sessions of bleomycin TACE, 56% of size reduction was achieved, comparable to literature data [2, 6, 7].

Since one bleomycin TACE resulted only in a 28% volume reduction, significantly less than literature data [2, 6, 7], we hypothesize that previous TAE sessions decreased the treatment's potential by blocking the vessels. Further studies are needed to investigate this hypothesis, but bleomycin TACE should be considered if a giant liver hemangioma seems to be resistant to conventional TAE treatment.

Conclusion

Our present case shows that bleomycin TACE can successfully decrease the size of giant liver hemangiomas even after several unsuccessful TAE sessions.

Conflict of interests: None to declare.

Authors' contribution: Conceptualization DBH and AD; GM and DBH compiled all relevant information concerning the presented case and did the literature review. AD, PAD, OH, AZF, AB, DAK, GM and DBH evaluated the whole treatment of the patients. GM and DBH wrote and prepared the original draft; GM and DBH made the visualisation. AZF, AB, DAK, OH reviewed and edited the manuscript. All authors have read and agreed to the published version of the manuscript.

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

Informed consent: Informed consent was obtained from the participant included in this report.

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