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A giant cavernous hemangioma of the liver extending into the pelvis

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ABSTRACT

INTRODUCTION: Giant cavernous hemangiomas are the most common tumors of the liver, ocurring in up to 20% of the general population. Given their benign course, asymptomatic nature and slow growth rate, treatment is rarely indicated. The case presented herein is unique as it describes an uncommon presentation of this common tumor and the circumstances in which surgical treatment is beneficial.
PRESENTATION OF CASE: We present a case of a 66 year-old patient with prostate cancer referred for evaluation of a massive 37 cm giant liver hemangioma, extending into the pelvis and in the planned field of radiation for prostate cancer, exhibiting rapid growth, and associated with significant symptomatology. Given these clinical characteristics, the patient was offered surgery and underwent a left trisectionectomy with an uneventful recovery. The patient's symptoms resolved and he was able to complete radiation to the pelvis.
DISCUSSION: In the context of an unusual presentation, this case presentation reviews the typical clinical and imaging characteristics of giant liver hemangiomas and expands on the current indications for treatment, emphasizing the role of enucleation and resection for patients meeting appropriate indications.
CONCLUSION: Although liver hemangiomas are extremely common, surgical treatment is rarely required. With appropriate indications, and when enucleation is not feasible or ideal, major liver resection is a safe alternative approach with excellent outcomes when performed in the right setting.

1. Introduction

Hemangiomas are the most common tumors of the liver, occurring in up to 20% of the general population [1]. Cavernous liver hemangiomas (CLH) are congenital vascular malformations that grow by ectasia—rather than hypertrophy, hyperplasia or neoplasia—therefore resulting in microscopic, and often macroscopic, cavernous spaces. CLH range in size from a few millimeters to over 20 cm; those over 5 cm referred to as giant hemangiomas [2]. Typically, CLH are asymptomatic and found incidentally, but can occasionally produce symptoms, most commonly abdominal pain. Bleeding/rupture or symptoms derived from obstruction of surrounding organs are extremely rare [1,2]. The classic imaging characteristics of liver hemangiomas include progressive peripheral nodular enhancement on dynamic cross-sectional studies (CT or MRI) and delayed centripetal fill-in. Treatment is rarely indicated given their benign course, lack of associated symptomatology and slow growth rate. We report a case that is particularly interesting given the rare presentation of this fairly common tumor, and expand on indications for treatment and surgical decision-making.

2. Presentation of case

A 66-year-old man with history of prostate cancer was referred to the hepatobiliary service for evaluation of a symptomatic and growing giant liver mass extending within the pelvis, and in the planned field of radiation for the prostate cancer. The patient reported feeling enlarging abdominal girth for the past 12 months associated with early satiety and new onset of pain/discomfort. Abdominal exam revealed a non-tender firm intra-abdominal mass extending across the whole abdominal cavity. Laboratory work-up was not contributory except for a total bilirubin of 1.3 mg/dl, and hepatitis serologies and tumor markers were negative. Cross-sectional imaging on current presentation revealed a significant increase in the size of the mass from 1 year prior ($10 \times 21 \times 27$ cm), now measuring $10 \times 25 \times 37$ cm. On dynamic CT the mass exhibited progressive nodular enhancement; however it was very heterogeneous containing diffuse large unenhanced cystic components and lacked evidence of centripetal fill-in (Fig. 1). It involved the majority of the liver—sparing only the right posterior section, and extended down into the pelvis (Fig. 2). Given the tumor's growth and location (precluding initiation of radiation therapy) as well as its symptomatology, the patient was offered surgical treatment—resection.

Abbreviations: CLH, cavernous liver hemangiomas; CT, computed tomography; GH, giant hemangiomas.

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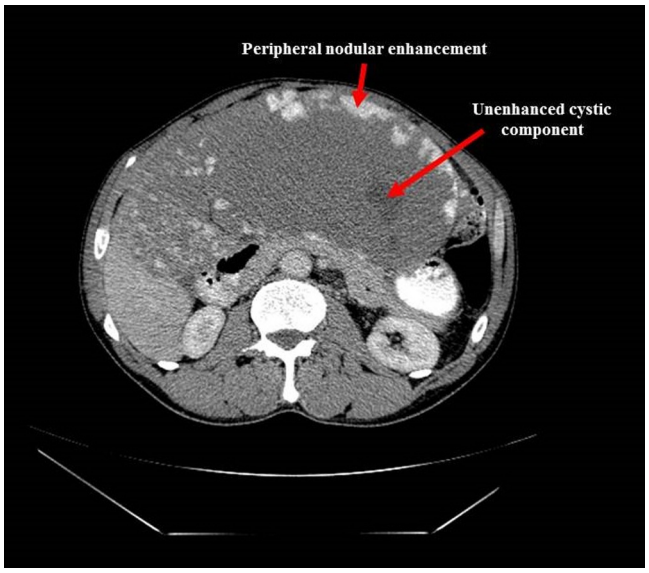

Fig. 1. Dynamic computed tomography (axial view) revealing a giant liver mass with peripheral nodular enhancement during venous/delayed phase—and a large unenhanced cystic component.

With the diagnosis of giant hemangioma (GH) established the goal of treatment was to remove the mass while preserving the majority of uninvolved liver. Preoperative imaging was not able to discern if clear planes of dissection would be present at the level of the hilum, although a completely replaced right hepatic artery was present. On exploration, despite the massive size of the GH, it was found to be pedunculated and free from the hilar structures allowing for safe dissection (Fig. 3). Early inflow control was obtained by transecting the proper hepatic artery (after confirming the replaced right hepatic artery was adequate), accomplishing downsizing of the tumor to 30 cm. Given the mass was encompassing the middle and left hepatic veins, and transection of these structures would be necessary for safe and complete resection, we opted for performing an anatomic resection rather than enucleation of the GH. The trunk of the middle and left hepatic veins was isolated and transected, in the form of an anatomic left trisectionectomy, although a significant

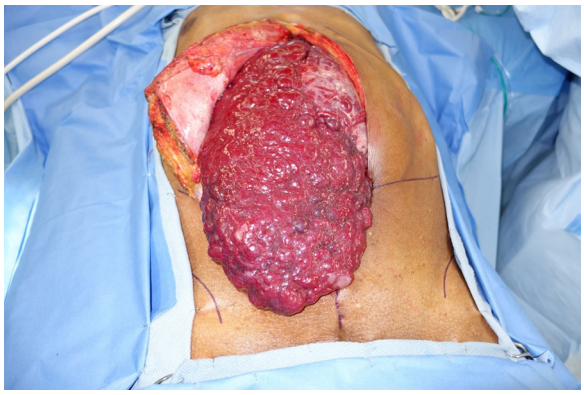

Fig. 3. View of giant liver hemangioma upon entering the abdomen. Note massive size as well as pedunculated feature allowing to eviscerate out of the wound, as it was free from hilar and other abdominal structures.

portion of uninvolved right liver parenchyma was preserved (Fig. 4). The patient recovered well and was discharged 4 days after surgery. He is now 9 months from resection and has completed radiation therapy. The preoperative symptoms of pain/discomfort and early satiety have fully resolved, with normalization of total bilirubin levels. Recent follow-up CT scan performed for prostate cancer surveillance revealed adequate hypertrophy of the liver and no evidence of residual or recurrent hemangioma (Fig. 5).

3. Discussion

We present a case of a patient with a massive giant hemangioma (GH) of the liver requiring surgical intervention. This case is particularly interesting as it highlights a number of uncommon circumstances not typically seen in patients with liver hemangiomas. First, the clinical presentation was not characteristic of a GH given its massive size with unusual extension into the pelvis, its rapid growth and the associated symptomatology. Further, despite typical progressive peripheral nodular enhancement on cross-sectional imaging, the common finding of delayed centripetal "fill-in" was absent [3], due to the presence of major cystic components. With these unusual clinical and imaging characteristics,

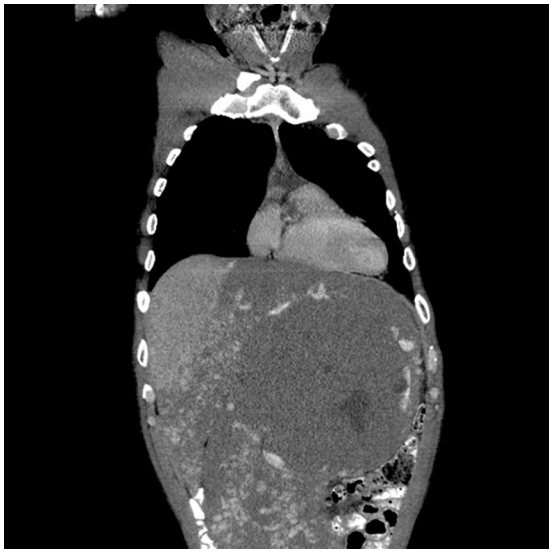

Fig. 2. Dynamic computed tomography (coronal view) in venous/delayed phase, revealing a giant heterogeneous liver mass extending from the diaphragm to the pubis, and involving the left lobe and right anterior section of the liver.

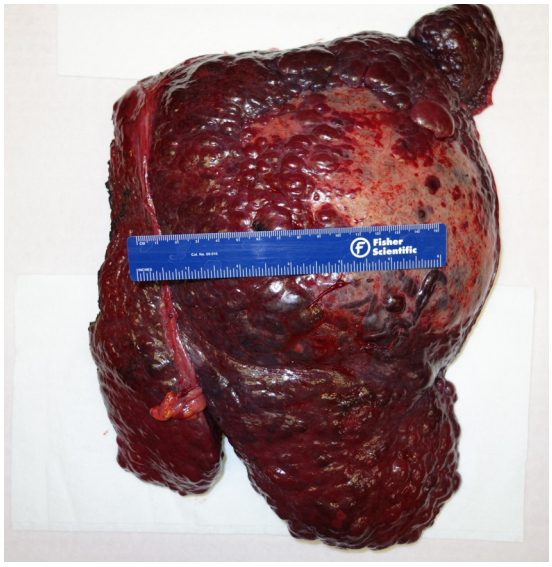

Fig. 4. Surgical specimen—left trisectionectomy, revealing a giant cavernous hemangioma measuring 30 cm (after arterial inflow control). The specimen weighed 2934 g.

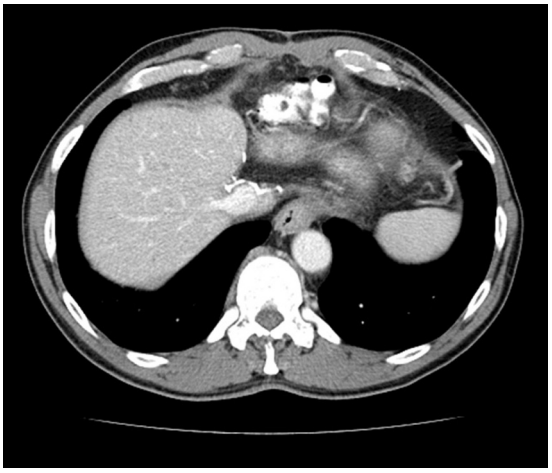

Fig. 5. CT scan 9 months after resection revealing staple line at the area of transection of the middle and left hepatic veins, and hypertrophied right posterior section of the liver.

the differential diagnosis was broader and included concern for an underlying neoplasia (i.e., hepatocellular carcinoma or cystadenoma/cystadenocarcinoma). Additional clinical, imaging and laboratory information were used to narrow the diagnosis so that appropriate treatment could be established; in the context of no underlying liver disease, with the pattern of peripheral nodular and progressive enhancement of a solitary lesion, normal tumor markers and lack of any distant disease, the preoperative diagnosis of a giant CLH was confirmed—with the cystic components representing large cavities with old blood/thrombus from the cavernous process and intermittent intralesional bleeding, further ruling out other more malignant processes.

The vast majority of CLH including giant hemangiomas (GH) have a benign course and require no treatment. When surgery is performed, the most common indication in adults is pain, followed by other less common reasons including uncertainty of diagnosis, complications such as rupture/bleeding or Kasabach–Merrit syndrome, and rapid growth [4]. Following hemangioma resection, pain usually subsides in approximately 63–75% of patients [2,4], emphasizing the importance of completing a comprehensive work-up for other causes of pain before considering surgical treatment. This case was also unique given the rapid growth rate observed. A recent study examining the natural course of liver hemangiomas found that only ~40% of CLH exhibit growth overtime, and for this select group, growth rates are in the order of 2 mm/year [5]. Our case had significant growth during a 1 year timespan, exceeding the reported growth rates, and stressing the previously reported association between larger GH and faster growth rates [5]. Although the faster growth rate observed in such cases is still poorly understood, it may be related to intralesional bleeding, as the cystic component with blood/thrombus represented the primary contributor of growth in our patient.

Treatment options for GH include arterial embolization and surgery for enucleation, resection or transplantation (rarely) [1,2,4]. A number of studies comparing the outcomes of resection and enucleation have found enucleation to be associated with lower risk of postoperative complications (primarily bile leak), and is currently the preferred approach [6]. Resection remains an appropriate alternative for cases in which the extent of the hemangioma is such that transection of major vascular structures is required and enucleation across these areas, although feasible is less safe and not ideal (as occurred in this case). In this setting, major liver resections including "super-extended resections" have been reported to be safe and associated with excellent outcomes when performed in high-volume centers [4,7].

4. Conclusion

Cavernous liver hemangiomas are the most common liver tumor, often presenting as giant hemangiomas (>5 cm). These tumors are typically asymptomatic and exhibit very slow growth rates. Treatment is rarely indicated. The case presented herein represents an ideal example of an uncommon presentation of this common tumor and the circumstances in which surgery is required. Almost invariably, surgery is only indicated for very large giant hemangiomas presenting with associated symptoms (most commonly abdominal pain), rapid growth, and/or when the diagnosis is uncertain (rarely). When the appropriate indications are present, enucleation is the preferred surgical approach. When enucleation is not ideal, major liver resection is a reasonable and safe alternative, with excellent outcomes when performed in the appropriate setting.

Conflict of interest

No conflicts of interest to report.

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Ethical approval

As this is a case-report of one individual (who has provided informed consent), an ethical approval was not required.

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.

Author contributions

Daniel Anaya conceived the initial idea of the study. Daniel Anaya and Jiayi Kong acquired data for publication, drafted manuscript and revised it critically for important intellectual content. All authors approved the final version of the manuscript submitted.

Research registry

This is a case-study – researchregistry300.

Guarantor

Dr. Daniel Anaya.

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References

[1] L.T. Hoekstra, M. Bieze, D. Erdogan, J.J. Roelofs, U.H. Beuers, T.M. van Gulik, Management of giant liver hemangiomas: an update, Expert Rev. Gastroenterol. Hepatol. 7 (2013) 263–268.
[2] M.S. Duxbury, O.J. Garden, Giant haemangioma of the liver: observation or resection, Dig. Surg. 27 (2010) 7–11.
[3] A. Bonder, N. Afdhal, Evaluation of liver lesions, Clin. Liver Dis. 16 (2012) 271–283.
[4] J.T. Miura, A. Amini, R. Schmocker, S. Nichols, D. Sukato, E.R. Winslow, et al., Surgical management of hepatic hemangiomas: a multi-institutional experience, HPB (Oxford) 16 (2014) 924–928.
[5] H.Y. Hasan, J.L. Hinshaw, E.J. Borman, A. Gegios, G. Leverson, E.R. Winslow, Assessing normal growth of hepatic hemangiomas during long-term follow-up, JAMA Surg. 149 (2014) 1266–1271.
[6] S.M. Lerner, J.R. Hiatt, J. Salamandra, P.W. Chen, D.G. Farmer, R.M. Ghobrial, et al., Giant cavernous liver hemangiomas: effect of operative approach on outcome, Arch. Surg. 139 (2004) 818–821.
[7] S. Pachera, H. Nishio, H. Yamada, Y. Yokoyama, T. Ebata, T. Igami, et al., Superextended hepatectomy for resection of multiple giant hemangiomas: report of a case, Surg. Today 39 (2009) 452–455.