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Torsion of a giant pedunculated liver hemangioma: Case report

Llener Castañeda Puicón^{a,b}, Yeray Trujillo Loli^{a,b,*}, Stefanie Campos Medina^c^a Department of Surgery, Hospital Nacional Daniel Alcides Carrión, Av. Guardia Chalaca 2176, Bellavista, 07016, Callao, Peru^b Universidad Nacional Mayor de San Marcos, Av. Grau Block 7, 15001, Lima, Peru^c Universidad San Martín de Porres, Alameda del Corregidor 1531, La Molina, 15024, Lima, Peru

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ABSTRACT

INTRODUCTION: Hepatic hemangioma (HH) is the most common benign neoplasm affecting the liver. Most cases are asymptomatic and incidentally found at imaging studies. Giant hepatic hemangiomas are uncommon, but pedunculated giant hemangiomas are even rarer, making their clinical and imaging diagnosis a challenge for the surgeon.

PRESENTATION OF CASE: Herein; we report the case of a 26-year-old male patient, who presented to the emergency ward with acute abdominal pain associated with a palpable mass, mimicking a complicated appendicular plastron, additionally were found it liver enzymes elevated. Then the patient was taken the operating room. An exophytic pedunculated liver tumor that projected towards the right iliac fossa was identified. It had undergone torsion on its pedicle. Transfixing hemostatic sutures were placed, proceeding with its exeresis. The patient tolerated the procedure and was discharged without complications on the third postoperative day. Pathological anatomy confirmed the diagnosis.

DISCUSSION: Giant pedunculated HH is an infrequent entity that generates mass effect on intra-abdominal organs causing pain depending on its location. In our case, HH size was 13 cm with elevated liver enzymes, only previously reported in one case of a 20-cm hemangioma. Surgery is indicated for cases of giant and symptomatic lesions.

CONCLUSION: Torsioned pedunculated giant liver hemangioma may be part of the differential diagnosis of a patient with acute appendicitis with elevated liver enzymes. The elevation of liver enzymes may be related to the size of the HH.

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1. Introduction

Hepatic hemangioma (HH) is the most common benign neoplasm affecting the liver. Most cases are asymptomatic and incidentally found at imaging studies. Its frequency of appearance ranges from 0.4 to 20% [1–3].

Liver hemangioma has female predominance due to its rapid growth in pregnancy, mainly associated to estrogen influence, and is commonly diagnosed in the 40–60 s. The right lobe is the most affected, there is no predominant racial factor and no risk of malignant degeneration has been reported [4,5].

Literature review shows that hemangiomas larger than 10 cm in diameter are considered giant hemangiomas [6], although sizes up to 20 cm have been reported [7]. Torsioned pedunculated liver hemangioma is a rare condition with a difficult diagnosis, its incidence is unknown and only a few case reports exist to date. This work is being reported in line with the SCARE 2018 criteria [8].

2. Case presentation

A previously healthy, 20-year-old man is admitted to the emergency ward with a 3-day history of abdominal pain that initiates in the epigastrium with migration of the pain to the right iliac fossa, which intensifies in spite of pain relievers, followed by nausea and hyporexia.

Physical examination reveals an asymmetric abdomen with a palpable mass projecting from the right iliac fossa to the hypogastrium and mesogastrium, painful on palpation and with positive peritoneal signs Mc Burney and Blumberg, and with a temperature of 38.5 °C. The rest of the systemic examination showed no alterations.

Laboratory was as follows; hemoglobin 15 g/dl, total leukocytes count 16,000/mL, with 1% bands, platelets 198,000/mL. Liver function test showed TGO 420 IU/l, TGP: 1521 IU/l, alkaline phosphatase 1304 IU/l, prothrombin Time 12.9 IU/l, INR: 1.1. The Alvarado's score of the patient was 9.

Contrast-enhanced abdominal tomography reports a heterogeneous 12 × 12 × 11 cm mass, with peripheral inflammatory changes in the right iliac fossa compatible with appendicular plastron or tumor mass. A second-opinion review of the CT images, after the

* Corresponding author at: Department of Surgery, Daniel Alcides Carrión National Hospital, Av. Guardia Chalaca 2176, Bellavista, 07016, Callao, Peru.
E-mail address: ytrujillo@unmsm.edu.pe (Y. Trujillo Loli).

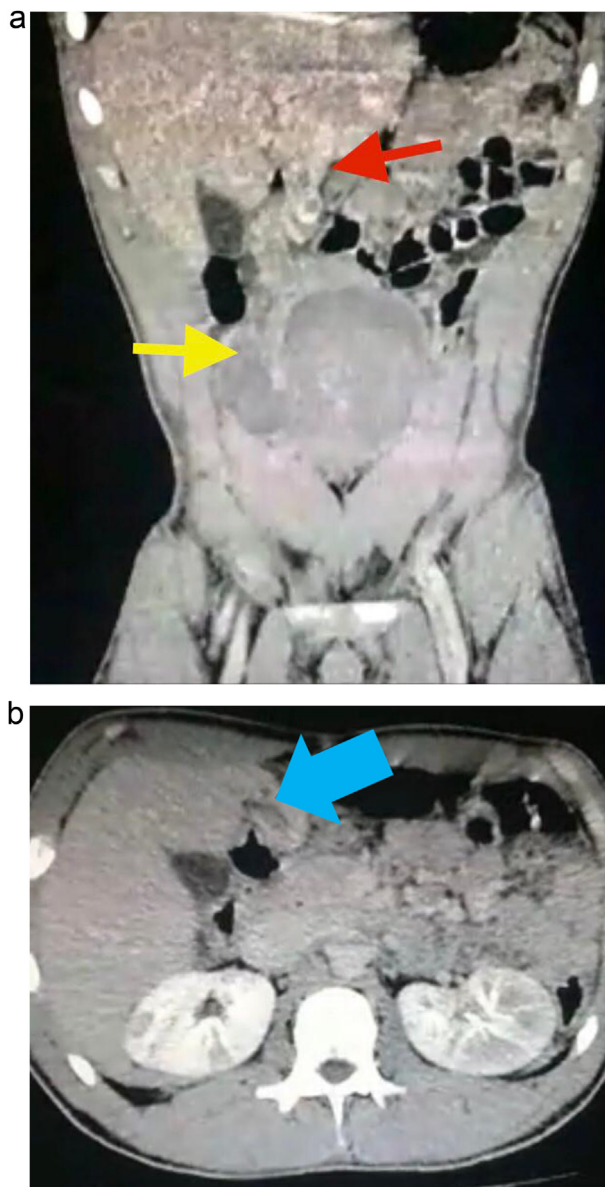


Fig. 1. Contrast-enhanced abdominal CT scan. **a** Coronal section: giant intraperitoneal hemangioma (yellow arrow) with a vascularized pedicle originating from segment IV (red arrow). **b** Axial section: shows the pedicle dependent on hepatic IV-B segment, with scarce contrast enhancement and tortuosity like the whirlpool sign (blue arrow).

intraoperative finding, reports it as a Giant Hepatic Hemangioma (Fig. 1).

Patient entered the emergency operating room with an indication of acute surgical abdomen. An exploratory laparotomy was performed. An ovoid smooth reddish mass was identified in the right iliac fossa. Macroscopically, the mass measured 13 × 12 × 7 cm and showed great vascularity (Fig. 2) in continuity with the liver by a wide pedicle originating from segment IVB and caudally extending to the right iliac fossa. It had undergone torsion on its peduncle (Fig. 3). The tumor was detorsioned, the pedicle was clamped at its proximal and distal ends and was, finally, resected. Hemostasis of the proximal end was achieved with transfixing stitches using absorbable multifilament suture. Patient tolerated surgery and was discharged without complications on the 3rd postoperative day. Histologic assessment with hematoxylin-eosin staining confirmed a cavernous hepatic hemangioma. (Fig. 4). Written informed con-



Fig. 2. Cavernous Hepatic Hemangioma: Exophytic lesion with a vascularized appearance dependent on the liver with a wide pedicle originating from liver segment IV.

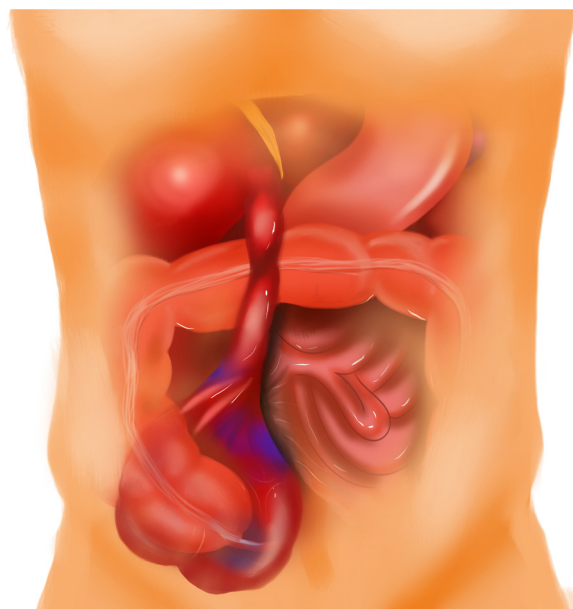


Fig. 3. Giant Hepatic Hemangioma: A twisted pedicle originating from hepatic segment IVB is seen.

sent was obtained from the patient for publication of this case report and accompanying images.

3. Discussion

Hepatic hemangioma (HH) is the most frequent tumor amongst benign liver lesions. Most are diagnosed incidentally and are asymptomatic [2]. On the other hand, one of its infrequent variants

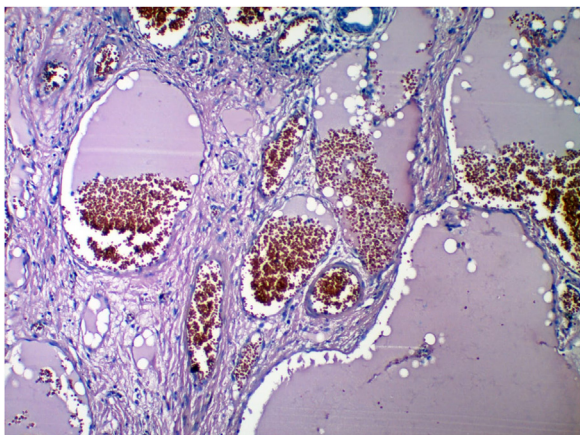


Fig. 4. Cavernous Hepatic Hemangioma: Proliferation of blood vessels of variable caliber with marked sinusoidal dilation and thinned walls covered by endothelium without atypia, occasionally filled with intraluminal thrombi over a hemorrhagic fibrous stroma.

includes pedunculated hepatic hemangioma; an exophytic lesion described only in few case reports. According to Al Farai A et al., only 24 cases have been reported to date worldwide [9]. It is frequently located in the left lobe, unlike our case report, where HH depends on the right lobe. Mocchiagiani et al. in their 7-year retrospective radiological study estimated a general prevalence of 10.9% for giant HH, of which only 0.47% were associated with rupture and bleeding, all of these cases being pedunculated hemangiomas [10]. These associations are compatible with the findings in this report.

HH can appear at any age, usually being diagnosed in the 40–60 s, predominantly in women, contrary to our case report that presents a 20-year-old male patient. In men, HH is frequently associated with coagulopathies such as Kasabach Merrit syndrome, a criterion that our patient does not meet [5].

Exophytic and extra hepatic growth of the hemangioma generates a mass effect on intra-abdominal organs producing pain [11]. The history of pain migration initiating at the epigastrium, as described in appendicitis, could also be a manifestation of pedunculated liver tumor, as experienced in our case. Complications of pedunculated hemangioma include torsion and infarction, producing mostly abdominal pain [12], as in our case, thus associated to intramural thrombosis, tumor inflammation, hemorrhage and rupture, as diagnosed intraoperatively in our patient.

Classification according to size is variable. Some authors consider lesions larger than 4 cm as giant hemangiomas [3]. Di Carlo et al., propose another classification based on a systematic review, in which HH with a diameter < 3 cm is considered as small, suggesting surgical resection for those larger than 10 cm [6]. In our case, a mass of these dimensions (13 cm × 12 cm × 7 cm), as identified in the intraoperative, is sufficient criteria to opt for resection as surgical behavior.

Imaging diagnosis is made using three-phase contrast abdominal tomography, observing well-defined and lobulated focal liver lesions, with globular peripheral enhancement in the portal-venous phase and centripetal filling in delayed phase [10]. In this case report, the torsion does not allow us to appreciate the typical HH images due to the lack of distal vascularization. Furthermore, a limitation in the reported case is the impossibility of establishing a certain radiological diagnosis in the preoperative period because the triphasic CT modality is not available in our hospital center.

In this case report, the preoperative diagnosis was plastronated acute appendicitis. Only 2 cases have been reported mimicking this surgical diagnosis [13,14]. HH can simulate several diagnoses like gastric submucosal and stromal cancer [11], less frequently colon

cancer, retroperitoneal tumors [15] and complicated hepatocellular carcinoma [16].

Surgery is indicated for cases of giant, symptomatic lesions, growth-prone masses, and in cases in which diagnosis is unclear and prone to complication. In 2016, Di Carlo I et al. estimated the risk of rupture of hepatic hemangioma at 3.2%, increasing when the lesion is pedunculated, peripheral and exophytic, with a statistically significant correlation between the size of mass, symptoms and risk of rupture [6]. In our case, surgical management was appropriate due to resection of HH.

To the best of our knowledge, our case is the second one reported with elevated liver enzymes, being the first reported by Al Farai A et al. in which the size of the hemangioma, 20 cm, can explain the elevation of liver enzymes.

4. Conclusion

Conclusively, torsioned pedunculated liver hemangioma is an infrequent pathology that implies difficulties in its diagnosis, so it should be included in the differential diagnosis of acute appendicitis, especially in a young patient with a palpable mass. The elevation of liver enzymes may be related to the size of the HH. The diagnosis is mainly done by imaging and surgery is the first line of treatment.

Declaration of Competing Interest

All authors declare that there is no financial or personal conflict of interest related to this work.

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Ethical approval

Ethical approval was exempted by our institution.

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 contribution

Castañeda Llenner: Writing – Original Draft. **Trujillo Yeray:** Writing – Review & Editing, Visualization, Supervision. **Campos Stefanie:** Writing – Review & Editing, Visualization.

Registration of research studies

Article submitted was a case report and therefore not applicable.

Guarantor

Llenner Castañeda Puicón (caedo.505@hotmail.com).

Provenance and peer review

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