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CASE REPORT

Case Report: Unusual presentation of concomitant gastric outlet obstruction and jaundice complicating giant hepatic angioma [version 1; peer review: 1 approved with reservations, 1 not approved]

Nour Elleuch , Aya Hammami , Amira Hassine, Wafa Ben Ameer, Wafa Dahmani, Ben Slama Aida, Ksiaa Mehdi, Braham Ahlem, Ajmi Salem, Hanen Jaziri, Jmaa Ali

University of Sousse, Faculty of medicine of Sousse Department of Gastroenterology, University Hospital Of Sahloul, Route de la Ceinture, Sousse, 4011, Tunisia

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Abstract

Hemangioma is the most common benign tumor affecting the liver. The vast majority of liver hemangiomas (LH) are less than 30 mm in diameter, asymptomatic, and are most often identified incidentally during radiological investigations for other reasons. Giant LH greater than 50 mm can lead to the development of symptoms and complications that require prompt surgical intervention. Herein, we report the case of a 58-year-old man who presented with gastric outlet obstruction and obstructive jaundice as a result of a giant hepatic hemangioma that was complicated with fatal spontaneous rupture.

Keywords

Giant hemangioma, Obstructive jaundice; gastric outlet obstruction; Acute spontaneous rupture

Open Peer Review

Approval Status

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20 Nov 2020	view	view

1. **Phillipe Abreu** , Santa Casa of Sao Paulo School of Medical Sciences, Sao Paulo, Brazil
University of Miami, Jackson Memorial Hospital, Miami, USA
Rushi Shah, University of Miami, Jackson Memorial Hospital, Miami, USA
2. **Shariful Islam** , San Fernando General Hospital, San Fernando, Trinidad and Tobago
University of the West Indies, Kingston, Jamaica

Any reports and responses or comments on the article can be found at the end of the article.

Corresponding author: Aya Hammami (aya_med@hotmail.fr)

Author roles: **Elleuch N:** Writing – Original Draft Preparation; **Hammami A:** Validation; **Hassine A:** Resources; **Ben Ameer W:** Methodology; **Dahmani W:** Visualization; **Aida BS:** Validation; **Mehdi K:** Supervision; **Ahlem B:** Conceptualization; **Salem A:** Visualization; **Jaziri H:** Software; **Ali J:** Supervision

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Introduction

Hemangiomas are the most common primary benign liver tumours with an incidence estimated up to 20%¹. Most of the lesions are less than 3 cm². They are typically asymptomatic and are most often discovered incidentally during radiological investigations for other reasons. Although there is no general consensus on size, the most authors agree that hemangiomas >4 cm are described as giant^{3,4}. Symptoms depend on the location and the size of the haemangioma. Concomitant gastric outlet obstruction (GOO) and obstructive jaundice is an extremely rare presentation of liver hemangiomas (LH)².

We report the case of giant LH developing in the left lobe of the liver responsible for GOO and obstructive jaundice with fatal outcomes secondary to spontaneous rupture.

Case report

A 58-year-old male Tunisian patient was admitted to our department owing to a 3-week history of worsening post-prandial vomiting, associated to abdominal pain in the right upper quadrant and jaundice. His past medical history was unremarkable.

The physical examination revealed a dehydrated patient with jaundice and moderately reduced general condition. His vital signs were stable, body temperature was 37.2°C, pulse rate was 72 beats/minute, and blood pressure was 120/75 mmHg. Abdominal palpation identified a large firm abdominal mass in the right upper quadrant extending to the epigastrium and the umbilicus.

Laboratory tests performed at admission showed hemoglobin levels of 12.6 g/dL, platelet count of 251,000/mL, international normalized ratio (INR) of prothrombin time of 1.12, albumin of 32 g/L (normal range: 36–48 g/L), aspartate aminotransferase of 45 UI/L (normal range up to 50 UI/L), alanine aminotransferase

of 65 UI/L (normal range up to 40 UI/L), alkaline phosphatase of 360 U/L (normal range: 32–91 U/L), gamma glutamyl transpeptidase of 310 UI/L (normal range up to 50 UI/L) and notable hyper bilirubinemia (total bilirubin of 279 µmol/L (normal range up to 17 µmol/L) and direct bilirubin 90 of µmol/L (normal range up to 7 µmol/L). Renal function tests, electrolytes and c-reactive protein level were all within normal limits.

As the patient presented with symptoms related to gastric outlet obstruction, upper digestive endoscopy was performed one week after admission, following fluid resuscitation and nasogastric decompression. It showed that the stomach was full of fluid and food particles with evident extrinsic compression of the prepyloric region and the duodenum. The abdominal CT-scan revealed a 24 x 18 x 32 cm lobulated mass with a dense contrast uptake. It caused compression of the stomach, the duodenum, the common bile duct, and the hepatic pedicle (**Figure 1**). A diagnosis of compressive giant LH was made on the basis of typical radiological aspects.

Although risky, surgery (partial liver resection) was decided upon because of the pressure being put on both the gastrointestinal system and the biliary tract. Unfortunately, before the intervention, the patient developed severe right upper quadrant abdominal pain, and became hemodynamically unstable. On palpation, the upper abdomen showed diffuse tenderness and fullness. Acute spontaneous rupture of the LH was strongly suspected and the patient died despite resuscitation attempts, three weeks after his admission.

Discussion

Hemangioma is a frequent benign hepatic tumor with an estimated incidence of 0.4 to 7.3% in the general population^{5,6}. Unfortunately, there are no national statistics for incidence in Tunisia. Regarding sex distribution, it seems that women are

Figure 1. Abdominal computed tomography images. A giant cavernous hemangioma, filling the left lobe of the liver, showing peripheral intermittent nodular contrast enhancement. with exophytic extension, compressing adjacent structures.

more frequently affected. The pathogenesis of this tumor is not yet completely understood. Female-specific hormones have been involved and it has been suggested that they stimulate the enlargement of haemangiomas⁷.

A diagnosis of LH can be made easily through imaging techniques including ultrasonography, CT, magnetic resonance imaging, either alone or in combination, with a high sensitivity greater than 90%. Occasionally positron emission tomography (PET) or angiography can be used⁸. On ultrasound, small hemangioma appears as a uniform hyperechoic mass with relatively well-defined margins. Larger lesions, can appear hypoechoic or inhomogeneous, because of possible necrosis, hemorrhage or fibrosis⁹.

LH occurs more frequently in the right lobe of the liver¹⁰. Although there is no general agreement on size, LH are considered giant when they exceed 40 mm in diameter³. However, other sizes have been subsequently specified to categorize a LH as giant (4, 8 and 10 cm in diameter)¹¹. Most lesions are less than 30 mm¹, usually asymptomatic, and most often diagnosed incidentally on imaging tests for other reasons, laparotomy or autopsy.

Giant hemangiomas are symptomatic in 90% of cases, with various signs and findings depending on the location and size of the lesion^{12,13}. The most frequently seen manifestation is abdominal pain resulting from the stretching of the Glisson capsule or necrosis of the lesion. In addition, giant LH may cause haematological abnormalities such as thrombocytopenia, anemia and leukopenia. Kasabach-Merritt syndrome is a life-threatening complication that may occur in 1.7% of patients with LH. It includes thrombocytopenia, consumptive coagulopathy and microangiopathic hemolytic anemia¹⁴. In rare cases, LH located in the left lobe, close to the hepatic hilum, may cause obstructive jaundice by biliary obstruction. GOO is also a rare manifestation of giant LH typically located in the left lobe of the liver, resulting from pressure on the stomach and the duodenum.

Spontaneous rupture of LH is an extremely rare and life-threatening event that is more likely to occur in lesions with a diameter ≥ 40 mm that are near the surface of the liver or with an exophytic growth¹⁵. It is associated with severe abdominal pain, peritonitis, and shock. The management of complications

is challenging as the mortality rate ranges between 60 and 75%. Only a few cases have been reported^{16,17}. Our patient had an extremely giant hemangioma exceeding 20 cm. To the best of our knowledge, this is the first case reported in literature to have LH with concomitant obstructive jaundice, GOO, and spontaneous rupture.

Treatment is only indicated when symptoms occur^{14,18}.

As most liver hemangiomas do not increase in size over time, it has been suggested that asymptomatic patients with hemangiomas less than 5 cm require no intervention or observation. Additionally, giant LH with no or few symptoms requires no treatment since there is little risk of developing complications.

Symptomatic LH is conventionally treated surgically. Enucleation and resection are the most commonly used techniques and they are both safe. If feasible, enucleation is generally the preferred approach¹⁹. The choice of approach depends on the experience of the surgeon and the location and size of the tumor. Other options, such as hepatic artery ligation, transcatheter arterial embolization or in rare cases liver transplantation, have also been proposed. Surgical management of extremely giant LH >20 cm remains challenging¹⁸. In the case of our patient, we considered that hepatic resection was the most suitable therapeutic option. Unfortunately, the patient died before surgery because of the occurrence of a fatal spontaneous LH rupture.

To conclude, giant LH does not necessarily require any treatment unless symptoms appear. Lesions close to the hepatic hilum may cause compression of the bile duct and cause obstructive jaundice, those close to the stomach and duodenum may cause GOO. The exact risk of spontaneous rupture remains unknown. The association of several complications such as the case of our patient is a very uncommon clinical presentation that requires prompt surgical intervention.

Data availability

All data underlying the results are available as part of the article and no additional source data are required.

Consent

Written informed consent for publication of their clinical details and clinical images was obtained from the family of the patient.

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Reviewer Report 05 July 2021

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Shariful Islam

¹ General Surgery, San Fernando General Hospital, San Fernando, Trinidad and Tobago

² Department of Clinical Surgical Science, University of the West Indies, Kingston, Jamaica

This is an unusual presentation of giant hepatic hemangioma.

The Title: use the commonly used term Hepatic hemangioma rather hepatic angioma.

Abstract: Needs further revision.

The authors should start by saying how rare the giant hemangioma is, the difficulties in the management and outcomes if not treated adequately or promptly - rather simply describing an hepatic hemangioma- which has been already described in the literature.

Introduction:

Should also include brief but updated treatment options.

Case report: Not written well.

Proper terminology was not used in many places. Needs further revision

Authors should explain the reasons for delayed treatment. Why an emergency surgery was not performed? and also why not early endovascular management was not offered to this patient.

Discussion:

Is too short, it should include the role of advanced endoscopic/endovascular option for a giant hemangioma

Conclusion:

Also requires further revision. In this section the authors need to highlight the importance of this case report to the literature.

Also to state what is learnt from this case report.

Decision:

In my opinion, it is an important finding and can add new information to the literature, however this article needs major revision, addressing all above concerns before indexing.

Is the background of the case's history and progression described in sufficient detail?

Yes

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?

Partly

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?

Partly

Is the case presented with sufficient detail to be useful for other practitioners?

Partly

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: All aspects of General and Laparoscopic and Breast Surgery

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Reviewer Report 17 May 2021

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Phillipe Abreu

¹ Department of Surgery, Division of Liver Surgery and Portal Hypertension, Santa Casa of Sao Paulo School of Medical Sciences, Sao Paulo, Brazil

² Department of Surgery, Miami Transplant Institute, University of Miami, Jackson Memorial Hospital, Miami, FL, USA

Rushi Shah

Department of Surgery, Miami Transplant Institute, University of Miami, Jackson Memorial Hospital, Miami, FL, USA

This is a very superficial case report from a patient who presented with a large hepatic hemangioma and was poorly managed, resulting in a fatal outcome.

Authors should better describe full information regarding the patient's clinical history and progression of the disease. Even in the course of the admission, it fails to describe symptoms and clinical presentation.

There is no discussion about the potential use of endovascular approach pre-surgical management of such large lesions, or any discussion about advanced endoscopic procedures that could be made to downsize the lesion prior to an exploratory surgical intervention was even considered (see attached citation as an example).

The current case as presented is a catastrophic example of how not to manage patients with liver hemangiomas, his clinical course is not even well described, rather only simple information as "died despite resuscitation attempts, three weeks after his admission". What resuscitation attempts? Why an emergency surgery was not performed? How come this patient remained in the hospital for 3 weeks and was not operated on before? Why no endovascular procedure was attempted?

Authors should better review this case in detail and present it in another way, considering alternatives of treatment and better justifications of the management adopted to clarify why the state of the art medicine was not the choice for the current patient.

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Is the background of the case's history and progression described in sufficient detail?

No

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?

Partly

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?

No

Is the case presented with sufficient detail to be useful for other practitioners?

No

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Abdominal Transplant Surgery, Liver and Pancreas Surgery

We confirm that we have read this submission and believe that we have an appropriate level

of expertise to state that we do not consider it to be of an acceptable scientific standard, for reasons outlined above.

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