Published online 2016 July 17.

Case Report

Torsion of a Giant Pedunculated Hemangioma of the Liver Presenting With Acute Abdomen: A Case Report

Aliasghar Darzi,¹ Hassan Taheri,² Sekineh Kamali Ahangar,³ Alameh Mirzapour Shafiei,⁴ and Yasser Asghari^{1,*}

Received 2016 April 04; Revised 2016 May 14; Accepted 2016 June 19.

Abstract

Introduction: Hemangioma is the most common benign tumor of the liver. Most cases are asymptomatic and do not require treatment. A hemangioma can rarely be pedunculated; as a result, it may undergo torsion and infarction, which can make it symptomatic.

Case Presentation: We report the case of a 45-year-old woman with acute abdominal pain due to torsion of a giant pedunculated hepatic hemangioma around its vascular stalk.

Conclusions: Pedunculated hemangioma of the liver is an uncommon benign tumor, a rare differential diagnosis for a mass located in the upper abdomen. All incidentally detected pedunculated hemangiomas must be surgically managed, as these have a tendency to become torsioned, and there is also a risk of malignancy or rupture.

Keywords: Torsion, Pedunculated, Hemangioma, Acute Abdomen

1. Introduction

Hemangioma is the most common benign tumor of the liver, with an autopsy prevalence of 0.4% - 20%. Women are more often affected, at a ratio of 5:1 - 6:1 (1, 2). Hemangiomas can occur at any age, are usually solitary, and are found more commonly in the right lobe, especially in the posterior segment (3). Liver hemangiomas are asymptomatic in most cases, and are commonly detected incidentally by imaging modalities such as ultrasonography, CT, or MRI (4).

Hepatic hemangiomas are usually < 3 cm in diameter, and those measuring > 4 cm are classified as giant (5). Giant hemangiomas can become large enough to cause symptoms due to mass effect on the abdominal viscera, abdominal pain, discomfort, hemorrhage, jaundice, and nausea (5-8). Exophytic forms of hepatic hemangiomas, especially pedunculated forms, are extremely rare, with only a few cases reported in the literature (9,10).

This case study is a report of a patient with a giant pedunculated hepatic hemangioma, who was referred to the emergency ward with acute abdominal pain due to torsion of the hemangioma around its vascular stalk.

2. Case Presentation

A 45-year-old woman with acute abdominal pain presented to the emergency ward of a private general hospital in Babol, northern Iran, on May 4, 2015. Her pain had originated in the epigastrium and had an incremental trend for two days prior to her admission. She had also experienced nausea, vomiting, and loss of appetite during that time.

The patient had a history of chronic abdominal discomfort and upper gastrointestinal bleeding (UGIB) due to gastritis. She also reported that she had taken oral contraceptive pills (OCPs) for approximately 5 years, many years earlier. On examination, her vital signs were all normal: blood pressure 125/75 mmHg, pulse rate 78 beats/min, respiratory rate 18 breaths/min, and oral temperature 37.3°C. Her abdomen was not distended on inspection. There was a mild generalized tenderness, as well as severe tenderness in the upper part of the abdomen, prominently the epigastrium, but there was no rebound tenderness or guarding. Complete blood count, basic metabolic profile, coagulation tests, and liver function tests were all within normal limits. B-HCG was negative.

There were no abnormal findings on the upright chest film and abdominal x-ray. Ultrasonography revealed a 92 \times 42 \times 72 mm solid ovoid mass with exophytic features,

¹Department of Surgery, Shahid Beheshti Hospital, Babol University of Medical Sciences, Babol, IR Iran

²Department of Gastroenterology, Ayatollah Rouhani Hospital, Babol University of Medical Sciences, Babol, IR Iran

³Clinical Research Department, Shahid Beheshti Hospital, Babol University of Medical Sciences, Babol, IR Iran

⁴Babol University Of Medical Sciences, Babol, IR Iran

^{*}Corresponding author: Yasser Asghari, Department of Surgery, Shahid Beheshti Hospital, Babol University of Medical Sciences, Babol, IR Iran. Tel: +98-1132256285, Fax: +98-1132256285, E-mail: bcrdc90@yahoo.com

originating from the anterior border of the left lobe of the liver. Abdominopelvic CT scan with contrast media demonstrated a 100×64 mm lobulated oval mass showing central necrosis in the anterior aspect of the abdomen, suggesting a mesenteric mass lesion, such as gastrointestinal stromal tumor (GIST), as the first differential diagnosis.

Due to her past history of UGIB, she underwent esophagogastroduodenoscopy by a gastroenterologist, and no mass or ulcer was observed. However, there was mass effect, most probably gastric compression due to an extragastric mass lesion, with a remote possibility of a submucosal lesion. Her colonoscopy was normal.

On her second day of admission, she had a low-grade fever and decreased hemoglobin (from 10 to 8.3 g/dL). According to the clinical and paraclinical findings, the unknown etiology of the mass, the possibility of malignancy, and the risks of bleeding and hemoperitoneum, we decided to perform surgical management.

In the operating room, under general anesthesia (GA) and with full monitoring in the supine position, exploratory laparotomy was performed. A large, multilobulated, dark red mass was discovered attached to the anterior edge of the left lobe of the liver by a vascular pedicle. It had undergone torsion on its pedicle. The mass was removed simply by legating the pedicle. Macroscopically, it measured 10.5 \times 8.8 \times 7 cm and weighed 316 g.

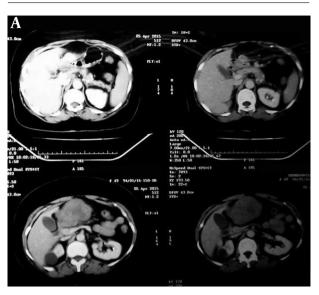
The histopathological analysis confirmed a huge pedunculated hepatic hemangioma, with marked vascular congestion and focal hemorrhagic infarct, with no evidence of atypia. The patient had an uneventful postoperative course, and was symptom-free. She was discharged from the hospital three days after surgery (Figures 1-3).

3. Discussion

Cavernous hemangioma is the most common benign tumor of the liver, probably with a congenital origin, and there is no potential for malignant transformation (1). It predominates in females 5:1 to 6:1, and is most common from age 30 to 50 years. It is usually discovered incidentally on imaging modalities such as ultrasonography, CT, and MRI (2).

These tumors can measure from a few millimeters up to 20 cm in size, and are commonly classified as giant if they measure > 4 cm in diameter (1). A pedunculated hemangioma can extend beyond the border of the liver (2), but giant pedunculated hemangiomas, as in the present case, are very rare (11-13).

Although most hemangiomas are asymptomatic, giant hemangiomas may cause abdominal discomfort. A good illustration of this is our patient, who experienced abdominal distention and a mass, abdominal pain, early satiety,



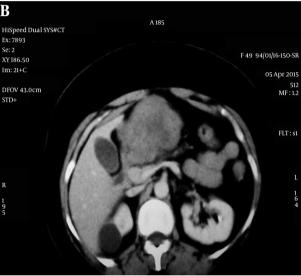


Figure 1. CT Scan of the Upper Abdomen Shows a Mass Between the Liver and Stomach, Probably From the Left Lobe of the Liver

nausea, vomiting, icterus, and thrombocytopenia (1, 6, 10, 14).

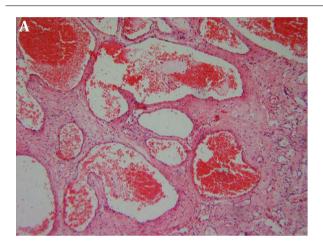
The first reported case of a giant pedunculated hepatic hemangioma was by Ellis et al. in 1985 (3). Another study reported a total of 24 cases of pedunculated, exophytic hemangiomas, which was consistent with the existing literature up to the year 2008 (2, 9).

Pedunculated hepatic hemangiomas are sometimes mistaken for other pedunculated tumors, such as hepatocellular carcinoma, hamartoma, focal nodular hyperplasia, and adenoma (3). Our patient had a history of OCP use, which could increase the incidence of adenoma. He-





Figure 2. A, Pedunculated hemangioma, torsioned Around its Stalk; B, Resected Hemangioma



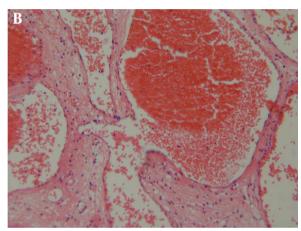


Figure 3. Histopathologically, the Lesion was Composed of Blood-Filled Vessels With Dilated Lumens and Thin Walls

mangiomas are usually small in size and asymptomatic by nature. Thus, follow-up is sufficient, without any treatment (2). However, if the hemangioma is large or giant and pedunculated, it can cause mass effect, abdominal symptoms, and a potential risk of rupture (2). Very rarely, these hemangiomas can spontaneously rupture with intra-abdominal hemorrhage, which may also occur due to blunt trauma, creating acute abdominal symptoms. For this condition, surgery is the best treatment choice, especially for a giant symptomatic hemangioma with an uncertain diagnosis.

A giant pedunculated hemangioma may undergo torsion around its pedicle, as in our case, and can become infracted, thereby becoming symptomatic. Pain is the most frequent symptom, which is due to infarction or pressure on the adjacent organs (1). Torsion of a giant pedunculated hemangioma is extremely rare, and had not been reported more than once in the literature up to the year 2010. That was a case in Turkey that mimicked acute appendicitis (1).

As the majority of hemangiomas are asymptomatic, they do not require treatment. In symptomatic cases, the treatment options are laparoscopy, open surgery, or angioembolization. The indications for surgical intervention include a palpable mass, rapid growth, thrombocytopenia, and rupture with intraperitoneal bleeding pain or epigastric discomfort, as demonstrated in our patient (4).

In the present case, we decided to perform surgical treatment after considering the symptoms, the large diameter of the mass, the inability to exclude malignancy, and the risk of rupture of the lesion. If surgery is contraindicated, other treatment modalities, such as radiation therapy, hepatic artery ligation, and arterial embolization can be used (10).

3.1. Conclusions

In conclusion, pedunculated hemangiomas of the liver are uncommon benign tumors that should be included in the differential diagnosis of any mass located in the upper abdomen. Imaging modalities, such as ultrasound, CT, MRI, radionuclide scintigraphy, and angiography, may be used to diagnose these tumors. This case report and other similar studies indicate that all incidentally detected pedunculated hemangiomas must be surgically managed, as they have a tendency to become torsioned.

Acknowledgments

We would like to thank the staff at the clinical research development center of Shahid Beheshti hospital in Babol for their cooperation.

Footnote

Authors' Contribution: Aliasghar Darzi performed the surgery and Hassan Taheri performed the esophagogastroduodenoscopy, Sekineh Kamali Ahangar and Alameh Mirzapour Shafiei collected the patient's information. The initial draft was written by Yasser Asghari, and Hassan Taheri revised it. All authors read and approved the final manuscript for publication.

References

- Ersoz F, Ozcan O, Toros AB, Culcu S, Bektas H, Sari S, et al. Torsion of a giant pedunculated liver hemangioma mimicking acute appendicitis: a case report. World J Emerg Surg. 2010;5:2. doi: 10.1186/1749-7922-5-2. [PubMed: 20148116].
- Ha CD, Kubomoto SM, Whetstone BM, Parascandola S. Pedunculated hepatic hemangiomas often misdiagnosed despite their typical findings. Open Surg J. 2013;7:1-5.

- Liang RJ, Chen CH, Chang YC, Hu RH, Sheu JC. Pedunculated hepatic hemangioma: report of two cases. J Formos Med Assoc. 2002;101(6):437-41. [PubMed: 12189652].
- 4. Chang CM, Lin SP, Lin CH, Hsu DW, Wei CK, Yin WY. Early Treatment for Symptomatic Giant Hepatic Hemangioma: Report of Three Cases and Literature Review. Surg J. 2007;2(4):45–9.
- Vivarelli M, Gazzotti F, D'Alessandro L, Pinna AD. Image of the month. Emergency presentation of a giant pedunculated liver haemangioma. *Dig Liver Dis.* 2010;42(6):456. doi: 10.1016/j.dld.2008.12.096. [PubMed: 19155194].
- Sohn MH. Evaluation of hepatic hemangioma by Tc-99m red blood cell hepatic blood pool scan. Korean J Nuclear Med. 2005;39(3):151-62.
- Bengisun U, Ozbas S, Gurel M, Ensari A. Laparoscopic hepatic wedge resection of hemangioma: report of two cases. *Langenbecks Arch Surg.* 2000;385(5):363–5. [PubMed: 11026709].
- Danet IM, Semelka RC, Braga L, Armao D, Woosley JT. Giant hemangioma of the liver: MR imaging characteristics in 24 patients. *Magn Reson Imaging*. 2003;21(2):95–101. [PubMed: 12670595].
- Masui T, Katayama M, Nakagawara M, Shimizu S, Kojima K. Exophytic giant cavernous hemangioma of the liver with growing tendency. Radiat Med. 2005;23(2):121–4. [PubMed: 15827530].
- Moon HK, Kim HS, Heo GM, Shin WG, Kim KH, Jang MK, et al. A case of pedunculated hepatic hemangioma mimicking submucosal tumor of the stomach. *Korean J Hepatol.* 2011;17(1):66-70. doi: 10.3350/kjhep.2011.17.1.66. [PubMed: 21494080].
- Bader TR, Braga L, Semelka RC. Exophytic benign tumors of the liver: appearance on MRI. Magn Reson Imaging. 2001;19(5):623-8. [PubMed: 11672619].
- Hosokawa A, Maeda T, Tateishi U, Satake M, Iwata R, Ojima H, et al. Hepatic hemangioma presenting atypical radiologic findings: a case report. *Radiat Med.* 2005;23(5):371-5. [PubMed: 16342910].
- Leone N, Saettone S, De Paolis P, Carucci P, Brunello F, De Angelis C, et al. Ectopic livers and related pathology: report of three cases of benign lesions. *Dig Dis Sci.* 2005;50(10):1818–22. doi: 10.1007/s10620-005-2944-7. [PubMed: 16187180].
- Biecker E, Fischer HP, Strunk H, Sauerbruch T. Benign hepatic tumours. Z Gastroenterol. 2003;41(2):191–200. doi: 10.1055/s-2003-37316. [PubMed: 12650132].