Spontaneous Rupture of Gallbladder Hemangioma Presenting as Hemoperitoneum

Satyaprakash Ray Choudhury, Mch¹, Sumit Subhadarshi Mohanty, MS¹, Premananda Pattanaik, Mch¹, Anasuya Lenka, MD², and Bhagaban Panda, MS¹

¹Department of Surgical Gastroenterology, Siksha O Anusandhan University, Institute of Medical Sciences and SUM Hospital, Bhubaneswar, India
²Department of Pathology, Siksha O Anusandhan University, Institute of Medical Sciences and SUM Hospital, Bhubaneswar, India

ABSTRACT

Hemangioma of the gallbladder is rare, with less than 10 cases reported in the literature. It may represent a hamartomatous proliferation of connective tissue in the gallbladder or may be congenital in origin. Although liver hemangiomas rarely present with spontaneous rupture, it has not been documented in gallbladder hemangiomas. This report presents a case of gallbladder hemangioma that ruptured spontaneously and presented with hemoperitoneum.

INTRODUCTION

Hemangioma of the gallbladder is rare, with less than 10 cases reported in the literature. It may present with mild upper abdominal pain or as an incidental radiological finding. When presenting as a gallbladder mass, it forms one of the differential diagnoses for carcinoma gallbladder.¹ Although spontaneous rupture of liver hemangioma is documented, ruptured gallbladder hemangioma has not been reported so far.² We describe a rare case of gallbladder hemangioma, which presented as an acute abdomen and shock due to spontaneous rupture.

CASE REPORT

A 62-year-old gentleman presented to the emergency department with acute abdominal pain. He denied any preceding trauma and did not have any comorbidities. At presentation, he was pale and hypotensive. The abdomen was distended with localized guarding in the right upper quadrant. Laboratory parameters revealed anemia with hemoglobin of 6.5 g/dL and elevated renal parameters (serum urea: 88 mg/dL, serum creatinine: 2.8 mg/dL) suggesting acute kidney injury. Liver function test was normal.

Ultrasonogram and noncontrast computed tomogram of the abdomen showed moderate hemoperitoneum and an ill-defined mass with recent hemorrhage abutting liver, gallbladder, and hepatic flexure (Figure 1). The patient was resuscitated with a working diagnosis of ruptured hemangioma of the liver. Angioembolization, although ideal, was deferred because of logistic and financial constraints.

The patient was taken for emergency surgery. Diagnostic laparoscopy revealed gross hemoperitoneum and a ruptured mass in the subhepatic region. A midline laparotomy was performed. After the evacuation of hematoma, a 12 × 8 cm mass arising from the anterior wall of the body of the gallbladder which had ruptured in its infero-medial aspect was seen (Figure 2). En bloc cholecystectomy was performed. The postoperative period was uneventful.

Histopathologic examination of the mass showed large cystically dilated thin-walled blood vessels arising from the subserosa of the gallbladder with evidence of hemorrhage. It also showed a breach of the capsule at an inferior aspect suggesting rupture. These features were consistent with cavernous hemangioma of the gallbladder (Figure 3).
DISCUSSION

Hemangioma is the most common neoplasm of the liver with a striking female preponderance. However, gallbladder hemangioma is extremely rare and literature shows only a few anecdotes. They are common in men and mostly asymptomatic. They may present as a gallbladder mass and mimic gallbladder carcinoma. Carcinoma gallbladder arises from mucosa unlike hemangioma, which is submucosal. Gallbladder carcinoma can present as subtle wall thickening, a polyp, or an infiltrating mass, whereas hemangioma is usually a well-defined mass with maintained mucosal integrity. Although classical imaging features and elevated tumor markers such as Carbohydrate antigen 19-9 (CA-19-9) and Carcinoembryonic antigen (CEA) can suggest malignancy, its absence does not rule out the latter. Hence, hemangioma of the gallbladder is approached with laparotomy and open cholecystectomy conventionally. Few reports of the laparoscopic approach in classical hemangioma have been described. The present case presented as an acute abdomen with shock and was promptly managed with emergency laparotomy because there were constraints for detailed imaging.

A liver hemangioma may present with spontaneous rupture in 1%–4% of cases. Hemangiomas on its inferior surface of the liver are more prone to rupture after trivial trauma because it could protrude out of the protective rib cage. However, no case of rupture and resultant hemoperitoneum has been reported in gallbladder hemangioma. The present case also mimicked the ruptured hemangioma of the liver. Only the intraoperative finding confirmed its origin from the gallbladder. The spontaneous rupture could be attributed to its large size. Angioembolization is the preferred modality of treatment for ruptured liver hemangioma. When the diagnosis is certain, the authors believe that the same treatment protocol can be applied for gallbladder hemangioma to stabilize the patient. Furthermore, cholecystectomy will eventually be mandatory because embolizing cystic artery may result in gallbladder gangrene. However, there is a lack of evidence because of its rarity.

Similar to hepatic hemangiomas, most of the hemangiomas of the gallbladder are a cavernous variety. The other types are venous and arteriovenous hemangiomas. The exact
pathophysiology has not been elucidated yet. The hamartomatous proliferation of connective tissue or congenital origin from embryonic mesodermal tissue in the subserosa of the gallbladder has been hypothesized. Our case is a type of cavernous variety that is composed of large cystically dilated thin-walled blood vessels in subserosa. The subserosal cavernous hemangiomas can attain a huge size and are predisposed for rupture similar to the present case scenario. Gallbladder hemangioma is an unusual condition diagnosed incidentally, although it may mimic malignancy warranting surgery. Our report of gallbladder hemangioma with spontaneous rupture managed with emergency laparotomy is the first of its kind in the literature.

DISCLOSURES
Author contributions: S. Ray Choudhury, SS Mohanty, P. Pattanaik, B. Panda wrote and approved the final manuscript. A. Lenka provided the pathology images and approved the final manuscript. SR Choudhury is the article guarantor.

Financial disclosure: None to report.

Informed consent was obtained for this case report.

Received April 30, 2020; Accepted September 4, 2020

REFERENCES
1. Funamizu N, Nakabayashi Y. Cavernous hemangioma of the gallbladder masquerading as a carcinoma. Case Rep Gastroenterol. 2019;13:219–24.
2. Cappellani A, Zanghi A, Di Vita M, Zanghi G, Tomarchio G, Petrillo G. Spontaneous rupture of a giant hemangioma of the liver. Ann Ital Chir. 2000; 71(3):379–83.
3. Ishak KG, Rabin L. Benign tumors of the liver. Med Clin North Am. 1975; 59(4):995–1013.
4. Crucitti A, La Greca A, Antinori A, Antonacci V, Magistrelli P. Cavernous hemangioma of the gallbladder. Case report and review of the literature. Tumori 2005;91:432–5.
5. Park JH, Lee JS, Choi GM, et al. Cavernous hemangioma of the gallbladder: A case report. iMRI. 2019;23:264–9.
6. Akama Y, Mizuguchi Y, Mamada Y, et al. A case of cavernous hemangioma of the gallbladder treated with single-incision laparoscopic cholecystectomy. Int Surg. 2016;101:431–6.
7. Adam YG, Huvos AG, Fortner JG. Giant hemangiomas of the liver. Ann Surg. 1970;172(2):239–45.
8. Suzuki H, Nimura Y, Kamiya J, et al. Preoperative transcatheter arterial embolisation for giant cavernous hemangioma of the liver with consumption coagulopathy. Am J Gastroenterol. 1997;92(4):688–91.
9. Jones WP, Keller FS, Odrezin GT, Kelly DR. Venous hemangioma of the gallbladder. Gastrointest Radiol 1987;12(4):319–21.

Copyright: © 2021 The Author(s). Published by Wolters Kluwer Health, Inc. on behalf of The American College of Gastroenterology. This is an open-access article distributed under the terms of the Creative Commons Attribution-Non Commercial-No Derivatives License 4.0 (CCBY-NC-ND), where it is permissible to download and share the work provided it is properly cited. The work cannot be changed in any way or used commercially without permission from the journal.