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CASE REPORT | BILIARY

Spontaneous Rupture of Gallbladder Hemangioma Presenting as Hemoperitoneum

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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).

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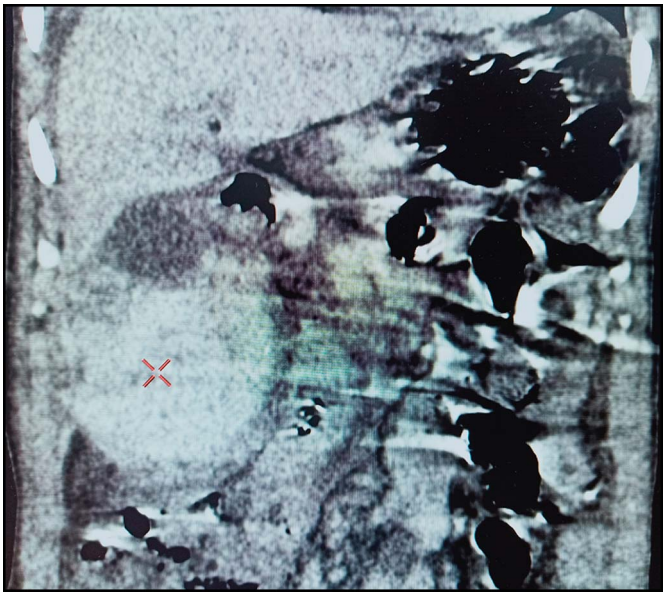

Figure 1. Noncontrast computed tomography scan, coronal view, showing a mass lesion probably arising from the liver and occupying the right subhepatic space with active hemorrhage.

DISCUSSION

Hemangioma is the most common neoplasm of the liver with a striking female preponderance.[3] However, gallbladder hemangioma is extremely rare and literature shows only a few anecdotes.[4,5] They are common in men and mostly asymptomatic. They may present as a gallbladder mass and mimic gallbladder carcinoma.[1] 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.[6] 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.[2] 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.[7,8] 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.

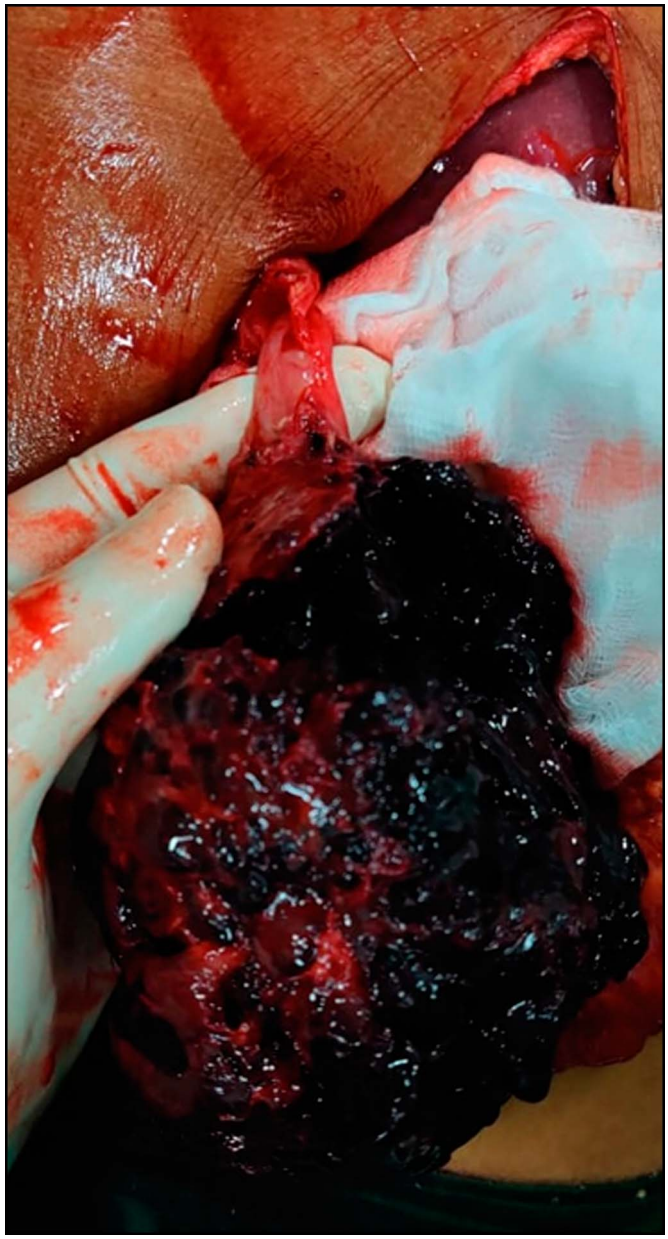

Figure 2. Intraoperative picture showing a large hemangioma arising from the body and fundus of the gallbladder, with breach in the capsule on inferomedial aspect.

Similar to hepatic hemangiomas, most of the hemangiomas of the gallbladder are a cavernous variety. The other types are venous and arteriovenous hemangiomas.[6,9] The exact

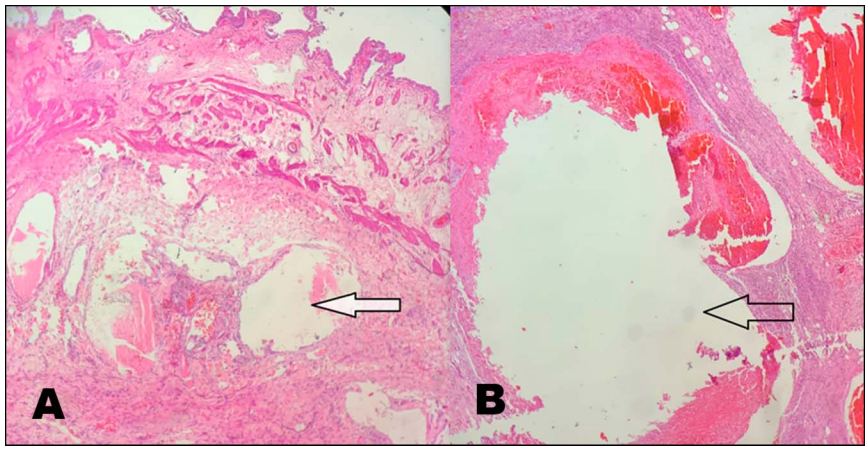

Figure 3. Hematoxylin and eosin staining at (A) 40× magnification and (B) 100× magnification showing the presence of cystically dilated thin-walled blood vessels (marked with arrows), predominantly in the subserosal layer, confirming cavernous hemangioma.

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.[9] 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.

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