




CASE REPORT

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A case of gallbladder hemangioma detected in a patient with jaundice and suspected Klatskin tumor: case report and review of the literature

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Abstract

Background and objective The diagnosis and treatment of benign tumors of the gallbladder and bile ducts are difficult due to their anatomical relationships with neighboring vital organs. Hemangiomas are non-epithelial benign tumors of the gallbladder. The gallbladder is an extremely rare localization for cavernous hemangiomas. To date, 7 cases of cavernous gallbladder hemangioma have been reported in the literature. Although it is seen very rarely, the main problem is that it mimics malignant lesions. Pre-operative diagnosis of gallbladder hemangiomas is difficult. Ultrasound (US), computed tomography (CT) magnetic resonance imaging (MRI), endoscopic ultrasound (EUS), and angiography are helpful in differential diagnosis.

Here, we aimed to present our case, which is the first case of cavernous gallbladder hemangioma and obstructive jaundice in the literature.

Case presentation A 49-year-old female patient was admitted with the complaint of pain in the right upper quadrant of her abdomen. Bilirubin levels were high due to obstructive jaundice. Abdominal CT and MRI showed an appearance in favor of hemangioma in the gallbladder. There was an increase in bile duct wall thickness on MRCP, and it was evaluated as suspicious for malignant neoplasia. The patient was operated on, and extrahepatic bile duct resection + Roux-en-Y hepaticojejunostomy procedure was performed. As a result of histopathology, hemangioma was detected in the gallbladder. There was no malignancy in the bile ducts.

Conclusion It should be kept in mind that the mass detected in the gallbladder in a patient with jaundice who is suspected of having a bile duct tumor may also be a hemangioma.

Keywords Gallbladder, Hemangioma, Treatment, Jaundice

Introduction

Diagnosis and treatment of benign tumors of the gallbladder and bile ducts reveal difficulties due to their anatomical relations with neighboring vital organs. Benign gallbladder tumors can mimic neoplasms developing in this area. Thus, it is important to know the characteristics of benign gallbladder lesions for differential diagnosis and treatment [1, 2]. Since hemangioma of the gallbladder is very rare, a rate related to its incidence has not

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been determined in the literature. Seven cases have been reported in the literature so far.

Here, a case of hemangioma of the gallbladder fundus, together with the obstructive jaundice clinical picture, which is an extremely rare association is presented.

Case

Patient information

A 49-year-old female patient, who was admitted to the emergency department with complaints of obstructive jaundice and intermittent abdominal pain for 4 months. The hypertensive patient had no previous operation history.

Clinical findings

Physical examination revealed tenderness in the right upper quadrant and epigastric region. The patient's sclera was icteric. There was a 5% weight loss in the last 6 months.

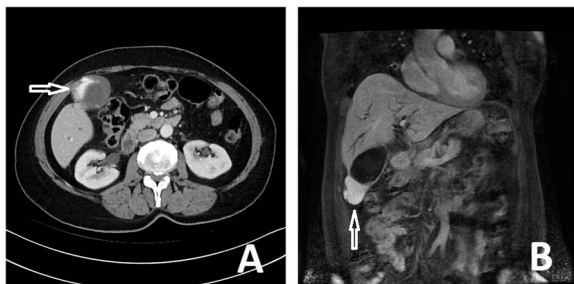


Fig. 1 Abdominal CT axial (A) and abdominal MR coronal (B) sections

Lab tests

Her liver function tests were high (total bilirubin 7.85, direct bilirubin 5.05, ALP 350). CA 19–9 levels were at the upper limit of normal and within the range of reference values.

Imaging studies

In the abdominal US, a mass with a diameter of 50×30 mm was observed in the gallbladder fundus. There were no pathological findings on the patient's chest CT. Subsequently, the mass with the size of 52×30 mm showing lateral extension on IV contrast-enhanced abdominal CT and MRI were evaluated in favor of gallbladder hemangioma (Fig. 1). MRCP showed dilatation in the intrahepatic bile ducts up to the hilus level, wall thickening, and focal diffusion restriction in the segment less than 1 cm at the common duct level were evaluated as suspicious for malignant neoplasia.

Surgical intervention

The patient was operated with the pre-diagnosis of Klat-skin tumor + gallbladder hemangioma. Extrahepatic biliary tract resection + Roux-en-Y hepaticojejunostomy procedure was performed.

Postoperative results

Gross pathology showed a well-defined cystic mass inseparable from the gallbladder fundus, 5×3 cm in size (Fig. 2). Microscopically, the tumor comprised of closely aggregated vessels lined by a single layer of flattened epithelium with no atypia. Immunohistochemically, the epithelium was positive for CD31 and CD34 and negative

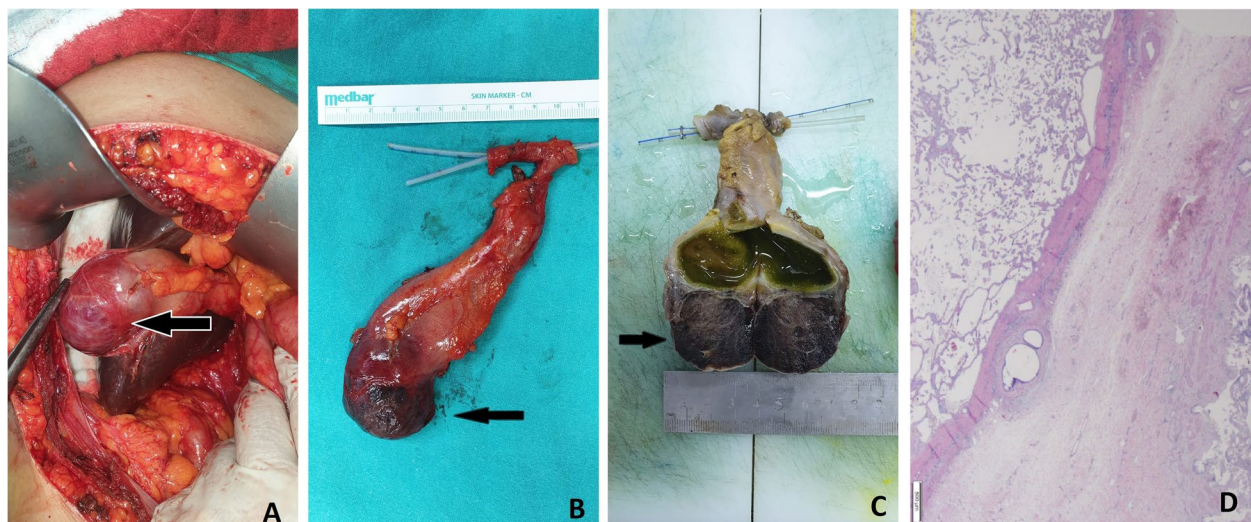


Fig. 2 Surgery picture of the gallbladder hemangioma (A). The picture of the specimen (B, C). Hemangioma located in the serosal aspect of gallbladder wall, hematoxylin-eosin ×2 (D)

for cytokeratin (AE1/AE3), which favored the diagnosis of gallbladder hemangioma (Fig. 2). The extrahepatic biliary duct wall showed mild fibrosis and chronic inflammation, and there was no atypia in the epithelium, which were all against the suspicion of Klatskin tumor. The patient was discharged on the fifth postoperative day and has been followed up for 4 months without any problem.

Discussion

The majority (88%) of gall bladder tumors are benign lesions. Most of them are adenomyomatosis, cholesterol polyps, and inflammatory polyps. Adenomyomatosis is most commonly located in the fundus and may be confused with malignant lesions of the gallbladder [2, 3].

Cavernous hemangiomas are most detected in the skin and liver. They constitute 73% of benign liver tumors. It is observed with a frequency of 0.4–7.3% in autopsy series [4]. Hemangiomas are non-epithelial benign tumors of the gallbladder [1]. The gallbladder is an extremely rare localization for cavernous hemangiomas. There is very little data in the literature regarding gallbladder hemangiomas. To date, 7 cases of cavernous gallbladder hemangioma have been reported in the literature. Although it is rare, the most critical problem is that it can be confused with malignant lesions. Differential diagnosis is vital in this regard. Gallbladder polyps can also become involved in diseases such as gallbladder tumors and xanthogranulomatous cholecystitis by thickening the gallbladder wall [3].

Histopathologically, hemangiomas are divided into 3 subtypes: capillary, cavernous, and arteriovenous. Within this classification, cavernous hemangiomas are aggressively growing tumors that do not tend to regress. Although they are benign, they may cause compression symptoms and rarely mortal hemorrhage, depending on their size [5].

Benign gallbladder lesions are usually asymptomatic. Although the cavernous hemangioma of our case originating from the gallbladder fundus reached 5×4 cm in size, there was no compression symptoms. Nevertheless, they may become symptomatic, depending on the size and location of the lesion. They may also present with acute biliary colic, epigastric pain, jaundice, and fever. They show similar symptoms to cholecystitis, choledocholithiasis, and gallbladder tumors and may be confused preoperatively with these diseases. A case of gallbladder hemangioma that was operated with the pre-diagnosis of a giant mesenteric cyst has been reported in the literature [6].

US is the first-line imaging technique of choice in the evaluation of gallbladder lesions. Considering its rapid rise in clinical practice, CT has also become the

mainstay in evaluating the gallbladder diseases. At the same time, MRI and EUS are also generally accepted as a problem-solving tool [7]. Despite the advances in endoscopic transpapillary biliary drainage, endoscopic biliary drainage, and cytology studies, there is no ideal radiological diagnostic method to distinguish benign and malignant lesions of the gallbladder. Angiography can also be used for differential diagnosis [8].

We found that the cases of cavernous gallbladder hemangioma reported in the literature were operated with the pre-diagnoses of gallbladder carcinoma, mesenteric cyst, cholecystitis, and hepatic angioma [5, 9]. No pre-operative correct diagnosis was made in any of these cases.

In our case, pre-diagnosis of gallbladder hemangioma was made during the abdominal CT and MRI examinations performed while investigating the etiology of jaundice, our hemangioma diagnosis was confirmed with laparotomy and postoperative histopathological examination. None of the gallbladder capillary hemangioma cases reported in the literature had gallstones. The patients most frequently applied with complaints of pain. Four patients had accompanying angiomatous changes such as vocal cord hemangioma, liver hemangioma, and intestinal hemangioma. Cholecystectomy or extended cholecystectomy operation was performed on all gallbladder hemangioma patients [5, 9]. In our patient, cholecystectomy and extrahepatic resection of the bile ducts were performed due to the presence of wall thickening, diffusion restriction, and malignancy suspicion in the common hepatic canal on MRI.

Conclusion

Gallbladder hemangioma is a rare entity, and USG, CT, MRI, EUS, and angiography are helpful in the diagnosis. Laparoscopic cholecystectomy should be the first option in cases if there is only hemangioma. The gallbladder should be removed in the endobag. If there is a risk of bleeding, suspicion of malignancy, or inability to reveal the anatomy, surgical exploration with laparotomy should be performed. It should be kept in mind that gallbladder hemangioma may be associated with obstructive jaundice as in our case.

Author contributions

F. Karahan: Concept, design and writing. A. Atay: Acquisition of data. N. Ekinçi: Critical revision for important intellectual content. O. N. Dilek: Drafting of the article. All authors read and approved the final manuscript.

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Declarations

Competing interests

The authors declare that they have no competing interests.

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