Cavernous Hemangioma of the Gallbladder Masquerading as a Carcinoma

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Keywords
Cavernous hemangioma · Gallbladder · Gallbladder tumor

Abstract
Cavernous hemangioma arising from the gallbladder is extremely rare. Here, we report a cavernous hemangioma of the gallbladder masquerading as a carcinoma. A 75-year-old man was referred to our institution for a follow-up study after gastrectomy. Abdominal computed tomography revealed that the gallbladder was filled with a low-density mass with calcification of the wall. The patient underwent extended cholecystectomy. Histologically, the tumor consisted of vascular endothelial cellular elements and hematomas. The postoperative course was uneventful without complications. Presently, only 7 cases of cavernous hemangioma of the gallbladder have been previously reported. This case serves as an important reminder to consider benign tumors including cavernous hemangioma when investigating all possible causes of a gallbladder tumor.
Introduction

Cavernous hemangioma (CH) is a congenital venous malformation that occurs in various organs containing vascular components. The most frequent locations of CHs are the liver and skin, including the face, mouth, and limbs. However, CH arising from the gallbladder is extremely rare [1]. Thus, we herein report the 8th case of CH occurring in the gallbladder masquerading as a carcinoma.

Case Report

Two years after distal gastrectomy for gastric cancer (Stage IA), a 74-year-old man presented to our institution for a regular follow-up study. A computed tomography (CT) scan of his abdomen revealed a mass in the gallbladder (Fig. 1a). For further investigation, contrast-enhanced CT and magnetic resonance imaging (MRI) were performed. The contrast-enhanced CT scan revealed a slightly enhanced region in a low-density area of the gallbladder (Fig. 1b). In addition, the MRI revealed a high-intensity region on T2- and diffusion-weighted images (Fig. 2a, b). Moreover, ultrasonography was performed, but the calcification of the gallbladder wall did not allow taking a clear view. Based on these findings, the patient was diagnosed with a gallbladder carcinoma. Therefore, the patient was admitted to our institute for surgical treatment. The results of the physical examination upon the patient’s arrival were as follows: blood pressure, 146/86 mm Hg; heart rate, 82 beats per minute; body temperature, 36.4°C; body weight, 86.0 kg; and height, 180.0 cm. The physical examination also revealed a lack of tenderness of the entire abdomen. The patient had no specific family or medical history excluding hypertension. Hematologic examination showed a white blood cell count of 4,000/µL (normal range: 3,500–9,000/µL), hemoglobin level of 10.8 g/dL (normal range: 14.0–16.0 g/dL), C-reactive protein level of 0.08 mg/dL (normal: ≤0.1 mg/dL), carcinoembryonic antigen level of 3.7 ng/mL (normal: ≤5.0 ng/mL), and cancer antigen 19-9 level of 13.6 IU/mL (normal: ≤37.0 IU/mL). The patient underwent extended cholecystectomy. During the surgery, the gallbladder wall appeared to be hardened and thickened and was strongly adherent to the duodenum, transverse colon, and bile duct. After dissection of the adhesions, the liver bed was resected using the Péan crushing method with a 2-cm margin from the gallbladder. In the resected specimen, the gallbladder was filled with a solid tumor and hematoma, which measured 10.0 × 8.0 × 5.5 cm (Fig. 3). Microscopically, the tumor comprised dilated and closely aggregated vessel components (Fig. 4a). Moreover, CD31 and CD34 staining revealed the presence of vessel components (Fig. 4b, c). The final histopathological diagnosis was CH of the gallbladder. The postoperative course was uneventful aside from mild ileus, and the patient was discharged 14 days after the surgery. The patient did not experience recurrence for 12 months after the surgery.

Discussion/Conclusion

The reported prevalence of gallbladder tumors varies from 3 to 7% in a healthy population [2]. Kwon et al. [3] reported that benign tumors accounted for 88% of gallbladder tumors.
The ability to diagnose gallbladder carcinoma has recently improved because of the developments of imaging modalities including CT, MRI, and endoscopic ultrasonography [4]. Recently, Itsuki et al. [5] reported that the diagnostic ability of bile juice cytology through endoscopic transpapillary gallbladder drainage was better than that through endoscopic bile duct drainage. However, there are still no radiological features that can be used to distinguish gallbladder carcinoma from other benign tumors [6]. Therefore, in cases of suspected gallbladder carcinomas, surgeons must perform surgical interventions for gallbladder tumors. To our knowledge, only 8 cases have been reported in the literature, including the present case. The previous reports are summarized in Table 1 [7–13]. Five (63%) patients had abdominal pain as a chief complaint because of local growth and bleeding of CHs. The median age of the patients was 53 years (range, 11–75 years), and cholecystolithiasis was observed in all cases. Three patients (38%) were diagnosed with gallbladder carcinomas and underwent surgical procedures. Thus, all patients received an accurate diagnosis of CH of the gallbladder from surgical intervention, including cholecystectomy and extended cholecystectomy. In conclusion, a CH arising from the gallbladder has rarely been reported. Therefore, surgical procedures are required to acquire an accurate diagnosis.

Statement of Ethics

All procedures followed have been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments. Informed consent was obtained from all patients for being included in the study.

Disclosure Statement

The authors declare that they have no conflict of interest.

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Author Contributions

N.F. wrote the initial draft of the manuscript. N.F. and Y.N. performed the operation. Y.N. contributed to collecting the data and assisted in the preparation of the manuscript. All authors approved the final version of the manuscript.
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Fig. 1. a Abdominal computed tomography (CT) revealed a low-density mass in the gallbladder. b Abdominal contrast-enhanced CT showed a slightly enhanced region of the tumor in the gallbladder (arrow).
Fig. 2. Magnetic resonance imaging on T2- (a) and diffusion-weighted images (b) showed a high-intensity area in the tumor (arrow).

Fig. 3. The resected specimen consisted of a 10-cm solid tumor with hematoma (arrow).

Fig. 4. Hematoxylin and eosin stain (×200 magnification) showed aggregated vessel components (a). CD31 (b) and CD34 (c) staining showed the presence of vessel components.
Table 1. Previously reported cases of cavernous hemangiomas of the gallbladder

| Case | First author [ref.] | Year | Age, years | Sex | Symptom   | Preoperative diagnosis | Operation methods         |
|------|----------------------|------|------------|-----|-----------|------------------------|---------------------------|
| 1    | Sewell [7]           | 1969 | 43         | M   | Epigastralgia | Benign tumor           | Cholecystectomy           |
| 2    | Moffat [8]           | 1973 | 57         | M   | Epigastralgia | Acute cholecystitis    | Cholecystectomy           |
| 3    | Cabrera [9]          | 1977 | 62         | M   | None       | Hepatic angioma        | Cholecystectomy           |
| 4    | Jones [10]           | 1987 | 11         | F   | Epigastralgia | CH of the liver        | Cholecystectomy           |
| 5    | Furukawa [11]        | 1997 | 56         | M   | Back pain  | Gallbladder carcinoma  | Extended cholecystectomy |
| 6    | Mayorga [12]         | 1997 | 50         | M   | Epigastralgia | Mesenteric cyst        | Cholecystectomy           |
| 7    | Crucitti [13]        | 2005 | 49         | F   | None       | Gallbladder carcinoma  | Cholecystectomy (L)       |
| 8    | Present case         | 2019 | 75         | M   | None       | Gallbladder carcinoma  | Extended cholecystectomy |

L, laparoscopy.