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Case Report

A Case of Cavernous Hemangioma of the Gallbladder Treated With Single-Incision Laparoscopic Cholecystectomy

Yuichi Akama[1], Yoshiaki Mizuguchi[1], Yasuhiro Mamada[1], Tetsuya Shimizu[1], Tomohiro Kanda[1], Yoshiharu Nakamura[1], Nobuhiko Taniai[1], Hideyuki Takata[1], Zenya Naito[2], Eiji Uchida[1]

[1]*Department of Surgery and* [2]*Department of Pathology, Nippon Medical School Hospital, Tokyo, Japan*

Although hepatic hemangiomas are common, gallbladder (GB) hemangiomas are extremely rare. We present a case of a cavernous GB hemangioma, which was detected during routine ultrasonography screening. The 51-year-old female patient was asymptomatic and had no relevant medical history. The preoperative imaging findings, including those of endoscopic ultrasonography, suggested that malignancy was highly unlikely in this submucosal tumor. Thus, we performed a single-incision laparoscopic cholecystectomy as a diagnostic and curative procedure. The postoperative pathological examination confirmed a GB hemangioma. In this case, the preoperative diagnosis was quite difficult, especially because of this rare presentation of a GB hemangioma. This is probably why the definitive diagnosis is established postoperatively in most previously reported cases of GB hemangioma.

Key words: Cholecystectomy – Endoscopic ultrasonography – Gallbladder – Hemangioma – Laparoscopic surgery – Single incision

Hemangiomas occur commonly in several organs, including the liver, brain, lung, and skeletal muscle. However, gallbladder (GB) hemangiomas are extremely rare.[1,2] In a PubMed search, we found only 8 other cases of GB hemangioma.[1,3–9] In general, it is quite difficult to make a definitive diagnosis of this rare condition. In addition, the definitive diagnosis of GB tumor can be quite difficult without pathological examinations, such as bile cytology. All of the previously reported cases of GB hemangiomas underwent surgery and a definitive diagnosis was made only after pathological examination of the specimen. Open laparotomy was performed in all cases because the possibility of

Corresponding author: Yoshiaki Mizuguchi, 1-1-5 Sendagi, Bunkyo-Ku, Tokyo, Japan.
Tel.: +81 3 3822 2131; Fax: +81 3 5685 0989; E-mail: yoshi1224@gmail.com

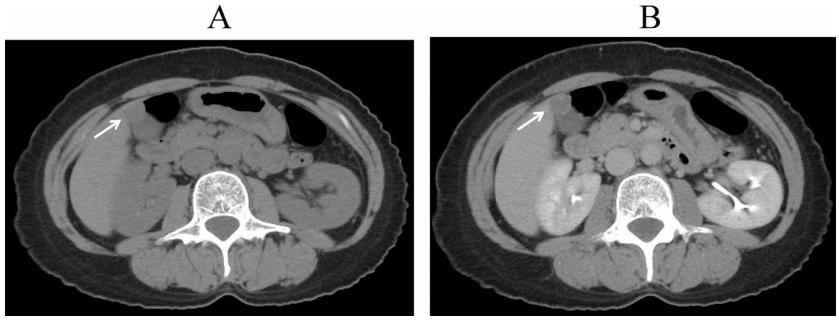

Fig. 1 Abdominal CT. (A) A slightly hyperdense lesion at the gallbladder fundus was visible by plain CT. (B) A few dots in the marginal area, which were enhanced by the contrast medium, were visible by contrast-enhanced CT.

malignancy could not be excluded based on preoperative imaging evaluations alone. In this study, we report a case of a cavernous GB hemangioma where the preoperative imaging findings, including those obtained by endoscopic ultrasonography (EUS) with Sonazoid (Daiichi Sankyo, Tokyo, Japan), excluded the possibility of malignancy, thereby allowing us to perform a single-incision laparoscopic cholecystectomy.

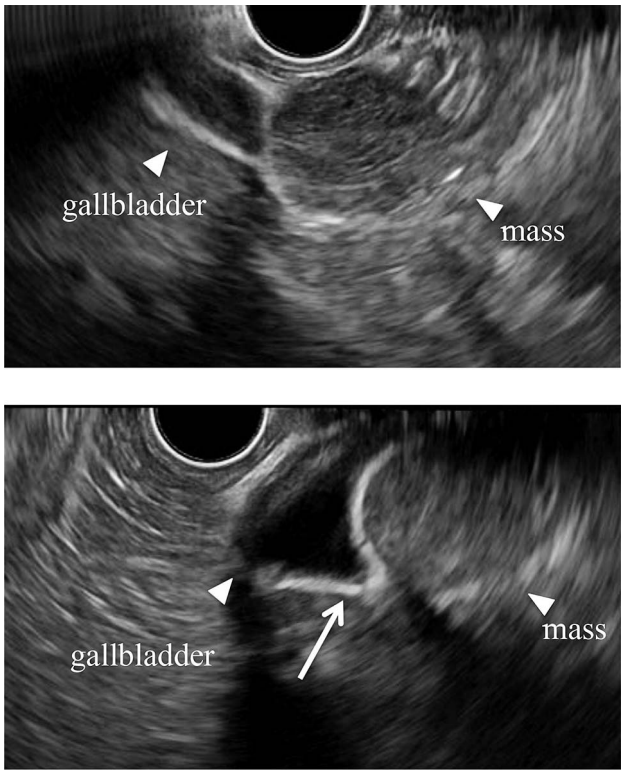

Fig. 2 Endoscopic US detected a well-defined, round, and homogeneous mass arising from the fundus of the gallbladder wall (upper). The surface of the mucosa was smooth and homogeneous without any thickened areas (lower).

Case Report

A 51-year-old woman was referred to our hospital for further evaluation because abdominal screening by ultrasonography (US) detected a GB mass. The patient was asymptomatic and had no medical history, except for cervical conization to treat cytological atypia.

The physical examination on admission was unremarkable. Laboratory data, including the hepatobiliary function and tumor marker levels, were within the normal limits: hemoglobin = 9.9 g/dL; total bilirubin level = 1.4 mg/dL; aspartate aminotransferase = 15 IU/L; alanine aminotransferase = 10 IU/L; and alkaline phosphatase = 143 IU/L. The tumor markers were also within the normal limits: carcinoembryonic antigen = 1.6 ng/mL; carbohydrate antigen 19-9 = 12.1 U/dL; and cancer antigen 125 = 24.1 U/mL.

Abdominal computed tomography (CT) detected a slightly hyperdense mass with a diameter of 17 mm at the GB fundus and a few dots in the marginal area, which were enhanced by the contrast medium, were likely to be Rokitansky–Ashoff sinuses (Fig. 1A, 1B). Transabdominal US identified a mass adjoining the GB fundus. Moreover, EUS detected the precise structure of a well-defined, round, and homogeneous mass arising from the GB fundus. The mucosal surface was smooth, intact, and homogeneous without thickened areas (Fig. 2). Endoscopic US with color Doppler and Sonazoid showed that blood flow was almost entirely absent from the mass, thereby indicating that the mass was not malignant. However, it was difficult to establish a definitive diagnosis based on the imaging findings alone, and thus adenomyomatosis or a submucosal tumor (e.g., gastrointestinal stromal tumor) was suspected. Therefore, we decided to perform a diagnostic operation by single-incision laparoscopic cholecystectomy. Intraoperatively, a dark-brown

Fig. 3 Intraoperative findings. Laparoscopy detected a dark-brown nodular lesion protruding from the gallbladder fundus and the mass was covered with the gallbladder serosa in a continuous manner.

nodular lesion was observed protruding from the GB fundus, which appeared to be covered with the GB serosa in a continuous manner. Part of it also adhered to the liver surface (Fig. 3). The mass and GB were resected with a portion of liver parenchyma. The operative specimen was a mass that measured 18 × 17 mm, which was located at the GB fundus and it originated from the GB wall (Fig. 4). The mucosal surface of the GB was intact, according to EUS. The pathological examination found large dilated submucosal vascular channels covered by a single layer of endothelium (Fig. 5). Because of its continuity with GB, the mass was diagnosed as a cavernous GB hemangioma and malignancy was excluded. The patient's postoperative course was uneventful. She was discharged on postoperative day 3 and her progress was favorable at the 1-year postoperative follow-up.

Discussion

In this study, we presented a case of GB hemangioma. Hemangioma is the most common benign mesenchymal tumor of the liver and its estimated prevalence ranges from 5 to 7%.[10] However, to the best of our knowledge, only 8 cases of GB hemangioma have been reported in the medical literature. The details of the 9 cases, including the present case, are listed in Table 1.[1,3–9] Arbab[1] reported the first case of a GB hemangioma in 1967. According to a review of 8 cases of GB hemangioma by Crucitti,[9] open laparotomy was performed in all cases, mainly because the possibility of malignancy could not be excluded based on the preoperative imaging findings alone.[7,9] As mentioned above, the very low possibility of malignancy according to our preoperative evaluation allowed us to perform a single-incision laparoscopic surgery in the present case. As far as we know, no other case has been reported in the medical literature where a GB hemangioma was excised by single-incision laparoscopic surgery. We consider that the assessment of the GB tumor by EUS was important in our case because the detection of a well-defined, round, and homogeneous mass without blood flow can exclude malignancy. None of the other case reports mentioned the use of EUS in the preoperative diagnosis. However, we obtained no findings to indicate malignancy in this case (e.g., infiltration of adjacent organ or heterogenic macroscopic appearance), but the operative strategy that we employed could be converted

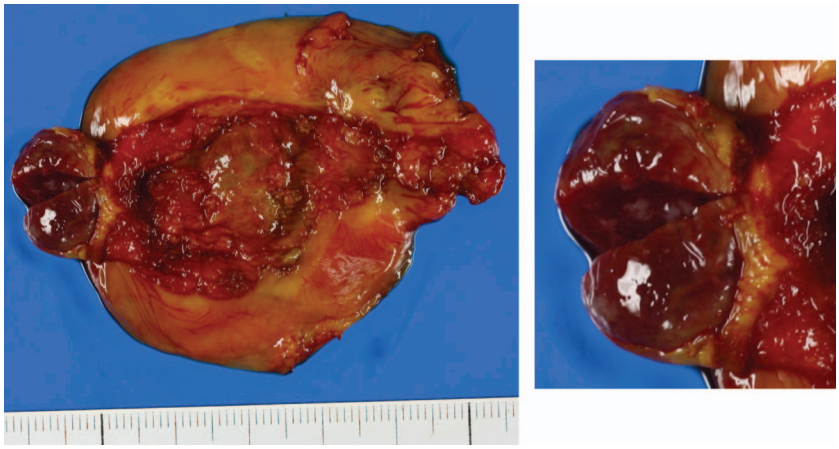

Fig. 4 Macroscopic findings based on the operative specimen. The surgical specimen was a mass measuring 18 × 17 mm, which was located at the gallbladder fundus and originated from the gallbladder wall. The serosa of the mass was continuous with that of the gallbladder.

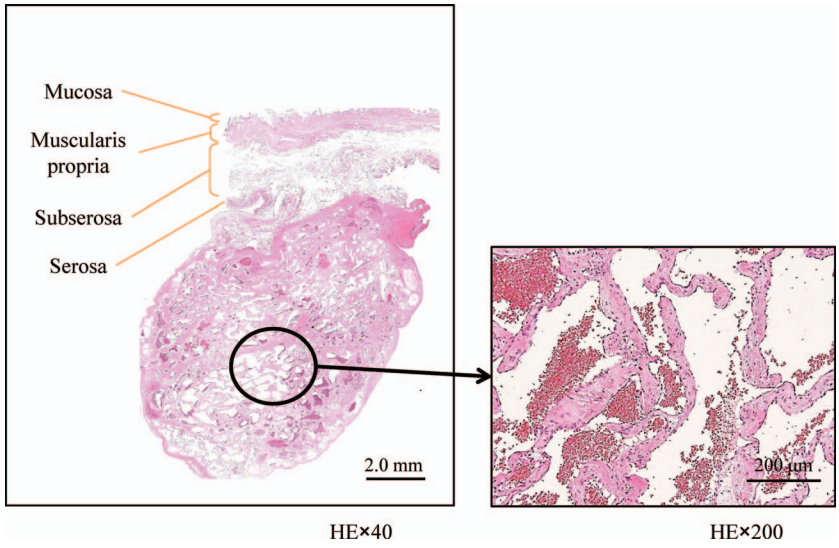

Fig. 5 Microscopy. The pathological examination detected large dilated submucosal vascular channels covered by a single layer of endothelium.

to multiple port laparoscopic surgery or open laparotomy. In addition, if a diagnosis of a GB malignant lesion is established postoperatively based on pathological examination, the patient might have had to undergo a second procedure with curative intent.[11]

Computed tomography might not be very useful for the definitive diagnosis of focal or diffuse thickenings of the GB,[12] while it may not be helpful for distinguishing benign and malignant tumors.[13] Although we were misled by the enhanced dots in the marginal area of the tumor according to contrast-enhanced CT, this could be a feature that is indicative of hemangiomas. Ultrasound is a noninvasive and relatively safe procedure. However, compared with transabdominal US, EUS has been reported to be an accurate imaging modality for evaluating GB,[14] and it may be very useful for assessing the mucosal surface and the depth of tumor invasion in the GB.[15,16] In this case, according

Table 1 Overview of previously reported cases of gallbladder hemangioma

Author	Age, sex	Examination	Preoperative diagnosis	Operation	Pathological diagnosis	Postoperative course
Arbab[1]	60, M	Intravenous cholangiography	Postcholecystectomy syndrome	Surgical excision	CH	Alive
Sewell[3]	43, M	X-ray, oral cholecystography	Benign tumor of the fundus	Cholecystectomy	CH	Alive
Moffat[4]	57, M	X-ray, oral cholecystography	Cholecystitis, cholelithiasis, choledocholithiasis	Subtotal cholecystectomy	CH	Alive
Cabrera[5]	62, M	Autopsy	Gallbladder adenoma	-	CH	Death by hypovolemic shock
Jones[6]	11, F	X-ray, US, arteriography	Cavernous hemangioma of liver	Cholecystectomy	VH	Alive
Furukawa[7]	56, M	CT	Gallbladder adenomyomatosis or adenocarcinoma	Extended cholecystectomy	Arterio-VH	Alive
Mayorga[8]	50, M	CT	Cystic mesenteric tumor	Cholecystectomy	CH	Alive
Crucitti[9]	49, F	AUS, CT	Gallbladder adenomyoma or adenocarcinoma	Cholecystectomy	CH	Alive
Present case	51, F	EUS, CT	Gallbladder adenomyomatosis or submucosal tumor	SILC	CH	Alive

AUS, abdominal ultrasonography; CH, cavernous hemangioma; VH, venous hemangioma.

to EUS, we considered that the mass was unlikely to be malignant based on our assessment of the mucosal surface and the properties of the GB mucosa. Color Doppler examination was not helpful for establishing a definitive diagnosis in this case because it did not detect blood flow within the hemangioma. Similar to our experience, a previous study reported that approximately 50% blood flow within a hemangioma could not be detected by color Doppler,[17] which might indicate that color Doppler cannot improve the accuracy of US. However, some previous reports suggest that the presence/absence of blood flow can be a useful sign for differentiating between benign and malignant tumors.[18,19] In addition, some reports state that contrast-enhanced US is a highly useful tool that can potentially help differentiate between benign and malignant tumors.[19–21] However, it is difficult to establish a preoperative diagnosis when such rare conditions affect the GB, including hemangiomas. In our case, EUS was helpful for excluding malignancy, which allowed us to perform a noninvasive and cosmetically appropriate diagnostic and curative procedure.

Conclusions

Although GB hemangiomas are benign tumors, a definitive preoperative diagnosis is difficult, probably because of their rarity. Endoscopic US is a potentially useful tool that can aid in their diagnosis and it may sometimes help exclude malignancies. However, further studies with a large number of patients are necessary to reach any definite conclusions.

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