CASE REPORT

Villous adenoma of gallbladder in a patient with systemic lupus erythematosus

Yuyun Xu, Jianhua Yuan, Vincent Chong,[1] Zhongxiang Ding

Department of Radiology, Zhejiang Provincial People's Hospital, Hangzhou, China, [1]Department of Diagnostic Imaging, National University Health System, Yong Loo Lin School of Medicine, National University of Singapore, Singapore

Villous adenomas occur most frequently in the rectum and colon. These tumors are rarely seen in the gallbladder. We report a case of gallbladder villous adenomas in a 69-year-old patient who has systemic lupus erythematosus (SLE). The patient was admitted for investigation of a gallbladder mass. Ultrasonography, computed tomography, and magnetic resonance imaging showed two well-circumscribed lobulated masses in the gallbladder. Open cholecystectomy was performed and histological examination revealed typical features of villous adenoma. This report describes the first case of villous adenomas of gallbladder with SLE, and documents its imaging findings comprehensively.

Key words: Computed tomography, gallbladder, magnetic resonance imaging, systemic lupus erythematosus, ultrasonography, villous adenoma

INTRODUCTION

Adenomas of gallbladder, the majority of cases associated with cholelithiasis, appear as polypoid structures. Gallbladder adenomas are uncommon, just found in 0.5% of the cholecystectomy, and villous adenomas of gallbladder have just been reported in a few papers.[1] Villous adenomas do not occur frequently outside the large bowel, with high potential for malignant transformation.[2] We report a case of gallbladder villous adenomas in a 69-year-old patient who has systemic lupus erythematosus (SLE).

CASE REPORT

A 69-year-old female patient was admitted in Zhejiang Provincial People's Hospital in 2011 for investigation of a gallbladder mass. She had a 15-year history of cholelithiasis (a gallstone measuring 2 cm diameter) detected by screening ultrasonography in a local hospital. No computed tomography (CT) examination had been performed before. She was also a known case of SLE diagnosed 10 years previously. Symptoms of SLE were controlled with methylprednisolone (8 mg/day) and azathioprine (100 mg/week).

On admission, her general condition was good with no history of fever, nausea, vomiting, epigastric pain, or weight loss. Tumor markers including AFP, CEA, CA125, CA19-9, CA15-3, and CA72-4 were within normal ranges, and blood chemistry tests revealed alanine aminotransferase (ALT) 22 μ/l (normal range 0-50 μ/l), aspartate aminotransferase (AST) 26 μ/l (normal range 10-52 μ/l), total bilirubin (TBIL) 16.2 μmol/l (normal range 3.4–24 μmol/L). White blood cells were 5.55×10^9/l, and blood sedimentation rate was 17.0 mm/h (normal <20 mm/h).

Abdominal ultrasonography showed two irregular iso-echoic masses within the lumen of the gallbladder, the bigger one's cross-section size was about 7.6 cm × 4.7 cm. There was no detectable extension through the gallbladder wall or invasion of adjacent structures [Figure 1]. CT showed two broad-based cauliflower-like masses in the gallbladder, from which the bigger mass was about 7.4 cm × 5.1 cm, and the smaller one 4.1 cm × 3.6 cm. The lesions measured 31-36 Hounsfield Unit on non-contrast CT and showed intense enhancement after administration of iodinated contrast material [Figure 2]. There was no evidence of extra-gallbladder extension or metastatic disease.

Magnetic resonance imaging (MRI) clearly identified the lesions. The mass in the body of the gallbladder was approximately 5.4×7.6×7.9 cm^3 while the lesion in the

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Address for correspondence: Dr. Zhongxiang Ding, Department of Radiology, Zhejiang Provincial People's Hospital, Hangzhou 310014, China. E-mail: hangzhoudzx73@126.com

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fundus measured about 4.0×4.0×4.5 cm^3. On T1-weighted images, the lesions were iso-intense to the liver, whereas on T2-weighted images, they showed high signal intensity [Figure 3]. A gallstone was noted in all imaging studies. The biliary system appeared normal and no associated hepatic lesions were detected.

Open cholecystectomy was performed and regional lymph nodes were removed. Histopathology revealed villous adenoma with regular arborization of the villo-tubular adenomatous epithelium. Most of the microscopic fields showed mild–moderate-grade epithelial dysplasia [Figure 4]. No cancerous epithelium was observed in specimens with hematoxylin–eosin staining. The regional lymphoid nodes showed chronic inflammatory reaction. After 17 months of follow-up, the patient reports no complaints.

DISCUSSION

Villous adenoma is a subtype of adenomatous polyps with malignant potential. Grossly, they appear as polypoidal masses with predominant villous elements. Adenomas of the gallbladder are uncommon and they are found in 0.5% of cholecystectomy specimens. They occur primarily in women, the majority of cases (50-65%) are associated with cholelithiasis, usually asymptomatic and discovered incidentally during a radiological evaluation for abdominal pain.[1] Villous adenomas are very rarely seen in the gallbladder. These lesions are most frequently in the rectum and colon.[2]

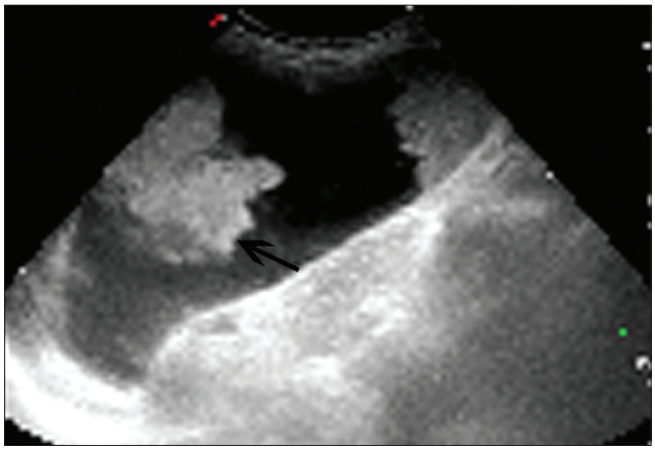

Figure 1: US shows intraluminal irregular masses (arrow). No extension through the gallbladder wall noted

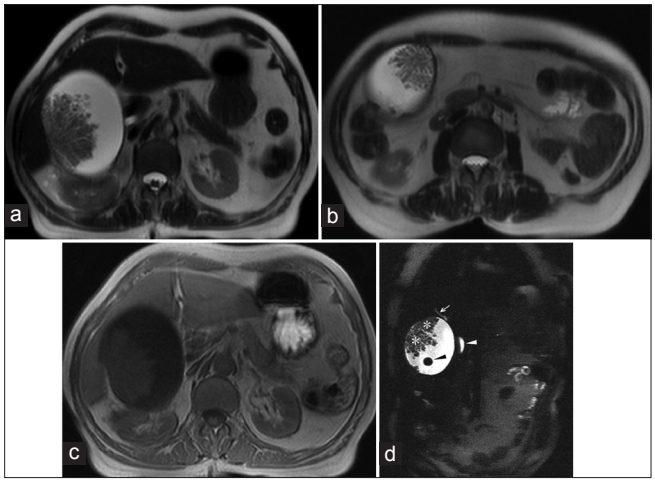

Figure 3: MRI shows frond-like intraluminal gallbladder lesions. Compared with liver, the masses display high signal on T2w image (a, b), and low signal on T1w image (c). Intrahepatic bile duct (white arrow) and common bile duct (white arrowhead) were not dilated on the thin-slab MRCP image (d). Note the villous adenoma (asterisk) and gallstone (black arrow) in the gallbladder

The histopathology and gross appearance of villous adenomas in the gallbladder and the gastrointestinal tract are similar. In 533 cases of cholecystectomy and 1300 randomly selected autopsies (mainly elderly individuals), villous adenomas were found in two resected cases (0.38%) and in one autopsy case (0.08%).[3] There are some reports of villous adenomas in the biliary tree and they are mostly located at the ampula of Vater.[4,5] Another two cases of gallbladder villous adenomas have been reported. One case which showed the lesion only by ultrasonography was in a patient with acromegaly,[6] and the other case was in a patient without other significant medical history, whose ultrasonography was negative and CT depicted the lesion.[7] These two villous adenomas were smaller than that in our report. In the current case, we had ultrasonography, CT, and even MRI as well as histology.

Villous adenoma is currently considered a premalignant condition. The following observations support the hypothesis of adenoma–carcinoma sequence of gallbladder carcinogenesis: (1) the presence of histological and genetic transition from adenoma to carcinoma, (2) close correlation between the size of the lesion and malignant

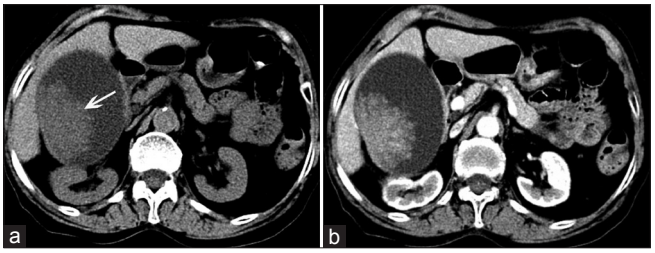

Figure 2: CT shows cauliflower-like broad-based lesions in the gallbladder. The mass is of intermediate density (about 31–36 HU) on non-enhanced CT (arrow) (a), with strong enhancement on contrast-enhanced CT (b)

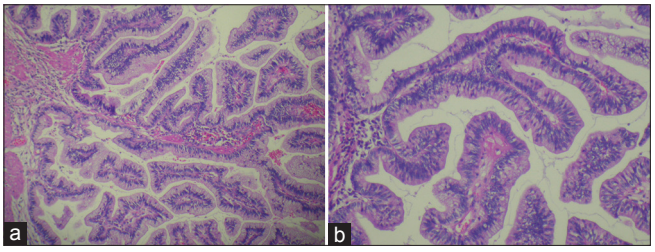

Figure 4: Microscopic examination demonstrated villous adenoma with mild to moderate atypical dysplasia (hematoxylin eosin staining, ×100 magnification in (a), and ×400 magnification in (b))

transformation, (3) orderly increase in age in the three groups, with a mean of 50 years for benign adenomas, 58 years for adenomas with malignant change, and 64 years for invasive carcinoma.[8,9] In a report of four patients with biliary villous adenomas (biopsy or surgically proven), two patients (50%) were subsequently found to have malignant lesions 3 years and 8 years following definitive diagnosis.[10] In addition, Kimura demonstrated positive immunohistochemical staining (CA19-9) in three patients. He also suggested that villous adenomas should be treated as a premalignant entity.[3]

As ultrasonography and CT technology improves, polypoid lesions are easily found. Ultrasonography takes advantage of depicting the gallbladder wall architecture exactly, but is easily interrupted by gas and too much fat. CT seems less sensitive in identifying the different layers of the wall. [11] MRI can show the surrounding structures better, and without ionizing radiation. The imaging characters of villous adenoma in the gallbladder are just like those in rectal and colon, duodenum, or bile ducts, showing soft tissue echo on ultrasonography, iso-hypodensity with various enhancing patterns on CT, while hypointense on the non-contrast T1-weighted images and heterogeneously hyperintense on T2-weighted images.[1,5,11,12] SLE is a multisystem autoimmune disease associated with significant morbidity and mortality. [13] The opinions about the relationship between SLE and the gallbladder disease are different. Some authors assume that the SLE patients are prone to biliary sludge and stone formation because of biliary dyskinesia like other collagen vascular diseases.[14] Other authors think gallbladder disease is no more common in SLE patients than that in the general population.[15] There are no previous reports on the relationship between gallbladder villous adenoma and SLE. The coexistence of SLE and gallbladder villous adenoma is thought to be probably coincidental in our case.

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