A unique case of gallbladder cancer arising in the neck side of segmental adenomyomatosis that mimicked hepatocellular carcinoma on contrast CT

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INTRODUCTION

Adenomyomatosis (ADM) of the gallbladder is defined as an epithelial proliferation and hypertrophy of the muscularis with outpouching of the mucosa into or through the thickened muscular layer, forming the so-called Rokitansky–Aschoff sinuses (RAS). Adenomyomatosis is categorized by its gross features as the diffuse type, fundal (localized) type or segmental type.1–3 The segmental type of ADM forms an annular stricture comprising a thickened wall dividing the gallbladder lumen into separate interconnected compartments, with or without diffuse wall thickening on the fundal side. Several reports have described that gallbladder cancer was sometimes found in the fundal side of the segmental type of ADM.1–3,5,6 But cancer arising on the neck side of the segmental type of ADM is extremely rare. We recently encountered a unique case of gallbladder cancer arising in the neck side of segmental ADM. Further, this case showed unusual morphology and mimicked hepatocellular carcinoma in an imaging study.

ABSTRACT

A 53-year-old Japanese female was admitted to our hospital for examination and treatment of an abdominal mass lesion. Contrast-enhanced abdominal computed tomography (CT) showed a well-circumscribed mass that measured approximately 5 cm. Although the tumor was located near the neck of the gallbladder, it seemed to be pedunculated from the liver parenchyma. From the enhancement pattern and morphological characteristics of the contrast CT images, the nodule was preoperatively diagnosed as hepatocellular carcinoma. The patient underwent a laparotomy, and it was intraoperatively revealed that the tumor existed solely in the neck of the gallbladder without serosal invasion. Whole-layer cholecystectomy with lymph node dissection was therefore performed. A pathological examination revealed that the papillary tumor filled up the neck side lumen of the segmental type of adenomyomatosis (ADM). The tumor was histologically diagnosed as papillary adenocarcinoma. Cases of papillary adenocarcinoma arising on the neck side lumen of the segmental type of ADM are extremely rare. Furthermore, no such case of gallbladder cancer which mimicked hepatocellular carcinoma has previously been reported.

Keywords: Gallbladder cancer, adenomyomatosis, papillary adenocarcinoma, hepatocellular carcinoma.
the diagnosis of acute cholecystitis. She underwent treatment for acute cholecystitis, and the presence of gallstones and an abdominal mass lesion were found by an imaging study. The patient was subsequently admitted to our hospital for further examination and treatment of the abdominal mass lesion. Laboratory tests performed on admission revealed no abnormality in the patient’s red blood cell count \((4.78 \times 10^6/\mu l)\), hemoglobin concentration \((12.2 \text{ g/dl})\), white blood cell counts \((7600/\mu l)\) or platelet count \((28.1 \times 10^4/\mu l)\). Serology and coagulation tests also showed no abnormalities. All of the examined tumor markers were within normal ranges, including those for carcinoembryonic antigen (CEA), carbohydrate antigen (CA)19-9, Dupan-2 antigen, SPan-1 antigen, protein induced by vitamin K absence (PIVKA)-II and alpha-fetoprotein (AFP).

Contrast-enhanced abdominal computed tomography (CT) showed a well-circumscribed mass that measured approximately 5 cm. The tumor showed mild enhancement in the early phase, and this enhancement was washed out in delayed phase (Fig. 1). Although the tumor was located near the neck of the gallbladder, it seemed to be pedunculated from the liver parenchyma of segment 6 (Fig. 2). The structures of the neck of the gallbladder and the cystic duct were unclear in the images. After discussion of whether the tumor originated from the liver parenchyma or the neck of the gallbladder, a preoperative diagnosis of hepatocellular carcinoma at segment 6 of the liver was made. Under the presumed diagnosis of hepatocellular carcinoma, surgical resection was performed. Intraoperatively, the tumor was found to be located at the neck of the gallbladder, and no serosal invasion or adhesion to other organs was observed. Whole-layer cholecystectomy was performed. The histology of the papillary adenocarcinoma and the surgical margin of the cystic duct were intraoperatively confirmed by a frozen section analysis. After the histological examination of frozen sections, lymph node dissection was additionally performed, and the operation was completed. The patient’s postoperative course was uneventful, and she was discharged from the hospital two weeks after the operation.

**PATHOLOGICAL FINDINGS**

A grossly papillary tumor filled up the neck side lumen of the segmental type of ADM (Fig. 3). The fundal side lumen contained white gall and a lot of gallstones. Histologically, atypical tumor cells proliferated as tubular structures in part, but dominantly showed a papillary structure, and were therefore diagnosed as papillary adenocarcinoma (Fig. 4a). The tumor invaded into the subserosal layer, but no serosal invasion was observed. A lot of dilated RAS were observed at the annular structure (Fig. 4b). No cancer metastasis was observed in the examined lymph nodes.

**DISCUSSION**

Adenomyomatosis of the gallbladder has not been considered to have malignant potential. However, there...
have been several reports suggesting that gallbladder cancer may originate from adenomyomatosis[6–10] or indicating that patients with the segmental type of adenomyomatosis have an increased risk of developing gallbladder cancer.[3,4] Most of these reported cases were of gallbladder cancer that arose on the fundal side of the segmental type of ADM. We have previously analyzed 97 cases of surgically resected gallbladder cancer and reported that 25 (25.8%) out of 97 cases were grossly accompanied with ADM.[5] In our previous series, there were only two cases where the gallbladder cancer had arisen on the neck side of the segmental type of ADM. Furthermore, no papillary adenocarcinoma was noted in our previous series of gallbladder cancer with ADM. Therefore, the present case where the papillary adenocarcinoma has arisen on the neck side of the segmental type of ADM is considered to be extremely rare.

Another interesting feature of present case is that the tumor mimicked hepatocellular carcinoma in the contrast CT images. Usually, it is easy to distinguish gallbladder cancer from hepatocellular carcinoma during imaging studies. We believe that there were several characteristics of the tumor and accidental factors that led to the unusual appearance of the nodule. First, the tumor filled up the cavity of the neck side of the segmental type of ADM and was demonstrated as a solid tumor in the CT images. Second, the lumen of the fundal side was markedly dilated, and made it look as if the lumen was the entire lumen of the gallbladder. Third, the tumor existed very close to the liver parenchyma, and it looked as if the tumor was pedunculated from the liver parenchyma. Fourth, the enhancement pattern of the contrast CT images was consistent with the pattern of HCC rather than gallbladder cancer. The usual enhancement pattern of invasive gallbladder cancer is a gradual enhancement reflecting the stromal component of the tumor. Usually, papillary adenocarcinoma contains a small amount of stroma, and the stroma of papillary adenocarcinoma contains a lot of blood vessels compared to other histological types of adenocarcinoma. These histological characteristics probably led to the early enhancement on contrast CT.

In summary, we have herein presented a unique case of gallbladder cancer arising in the neck side of a segmental adenomyomatosis that mimicked hepatocellular carcinoma on contrast CT. Although this is a very rare case, and was modified by several unusual conditions, we have reported this case as a reference for similar cases that might be encountered in the future.

**REFERENCES**