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CASE REPORT

Gallbladder sarcomatoid carcinoma: A case report and review of current literature



Ming-Wun Wong^a, Ming-Jen Chen^{a,b,c}, Chih-Jen Chen^{a,b,c},
Chien-Yuan Hung^{a,b,c}, Horng-Yuan Wang^{a,b,c},
Ching-Wei Chang^{a,b,c,d,*}

^a Division of Gastroenterology, Department of Internal Medicine, Mackay Memorial Hospital, Taipei, Taiwan

^b Mackay Junior College of Medicine, Nursing and Management, Taipei, Taiwan

^c Mackay Medical College, New Taipei, Taiwan

^d Institute of Traditional Medicine, National Yang-Ming University, Taipei, Taiwan

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KEYWORDS

Carcinoma;
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Therapy

Summary Gallbladder sarcomatoid carcinoma is a rare neoplasm characterized by the simultaneous presence of malignant epithelial and mesenchymal components. There is little information about the clinical behavior and optimal treatment for these tumors. The current study examines the case of a 52-year-old female patient who presented with postprandial epigastric pain that had persisted for 1 year. A diagnosis of gallbladder sarcomatoid carcinoma was confirmed by pathology after radical resection. Peritoneal metastasis occurred 3 months later, and the patient died 6 months after diagnosis. This study found that the prognosis of gallbladder sarcomatoid carcinoma is poor even after curative resection and there is as yet no known effective adjuvant therapy.

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Introduction

Adenocarcinoma is the most common type of malignant tumor generated in the gallbladder, whereas sarcomatoid

carcinoma (also called carcinosarcoma) is rare [1]. The diagnosis of sarcomatoid carcinoma is generally made pathologically and requires the simultaneous presence of both malignant epithelial and mesenchymal components. Immunohistochemically, the epithelial component is positive for cytokeratin and the mesenchymal component is positive for vimentin [2]. In a meta-analysis of sarcomatoid carcinoma, adenocarcinoma was found to be the most common pathology in the epithelial component, followed

* Corresponding author. Division of Gastroenterology, Department of Internal Medicine, Mackay Memorial Hospital, Number 92, Section 2, Chung-Shan North Road, Taipei 10449, Taiwan.
E-mail address: wei591026@gmail.com (C.-W. Chang).

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by mixed adenosquamous cell carcinoma and squamous cell carcinoma. In the mesenchymal component, spindle cell pathology occurred most frequently followed by combined spindle cell and chondroid features, and osteoid histology [3].

Sarcomatoid carcinoma of the gallbladder was first reported in 1907 by Landsteiner [4]. Since then, less than 100 further cases have been reported in the English literature [1]. Like gallbladder adenocarcinoma, gallbladder sarcomatoid carcinoma is most common in women. The female/male ratio is between 3.25:1 and 5:1 [1,3]. The age at diagnosis ranges from 45 to 91 years old [3]. Sarcomatoid carcinoma is rare at sites such as the lungs, kidneys, gastrointestinal tract and gallbladder, and occurs more commonly in the uterus where it is known as a malignant mixed Müllerian tumor [5].

Case report

A 52-year-old female patient presented with postprandial epigastric pain that had persisted for 1 year. The pain was mild and improved after antacid treatment. She was seen at our hospital after being diagnosed with a hepatic tumor at a local clinic. Abdominal ultrasonography revealed one mixed echogenic tumor approximately 9.5 cm from the gallbladder sac and extending to S4–5 of the liver. The common bile duct was dilated to 1.4 cm (Fig. 1). Liver function tests and total bilirubin levels were within the normal range. However, carbohydrate antigen 19-9 (CA 19-9) was elevated to 60.9 U/mL. A follow-up abdominal computed tomography (CT) confirmed a 7.5 cm sized gallbladder cancer with liver invasion (Fig. 2), consistent with clinical Stage III cancer according to the TNM system (tumor-node-metastasis classification system).

Cholecystectomy and segmental hepatectomy of segments S4b and S5 were performed. An 8 cm, friable gallbladder mass had grown outward and involved the liver (Fig. 3). Pathologically, the tumor was a sarcomatoid carcinoma in Stage III (Fig. 4). The surgical margin was free of vessel or cystic duct invasion. The tumor had an adenocarcinoma component with some glandular structures and showed positive staining for cytokeratin (AE1/AE3) and a sarcoma component with spindle cells staining positively for vimentin.

The postoperative CA 19-9 level decreased to 28.5 U/mL. Epigastric pain improved and the patient did not report any other discomfort. However, 3 months later, the follow-up abdominal CT revealed a peritoneal, poorly enhancing soft tissue mass over the anterior of the liver and variably sized nodules on the omentum (Fig. 5). A CT-guided biopsy confirmed metastatic peritoneal sarcomatoid carcinoma. The patient received chemotherapy consisting of three courses of cisplatin (30 mg/m²) and fluorouracil (2600 mg/m²). The patient died 6 months after diagnosis.

Discussion

We reported the case of a 52-year-old female patient who presented with postprandial epigastric pain that had persisted for 1 year. Peritoneal metastasis occurred 3 months later, and the patient died 6 months after diagnosis. The

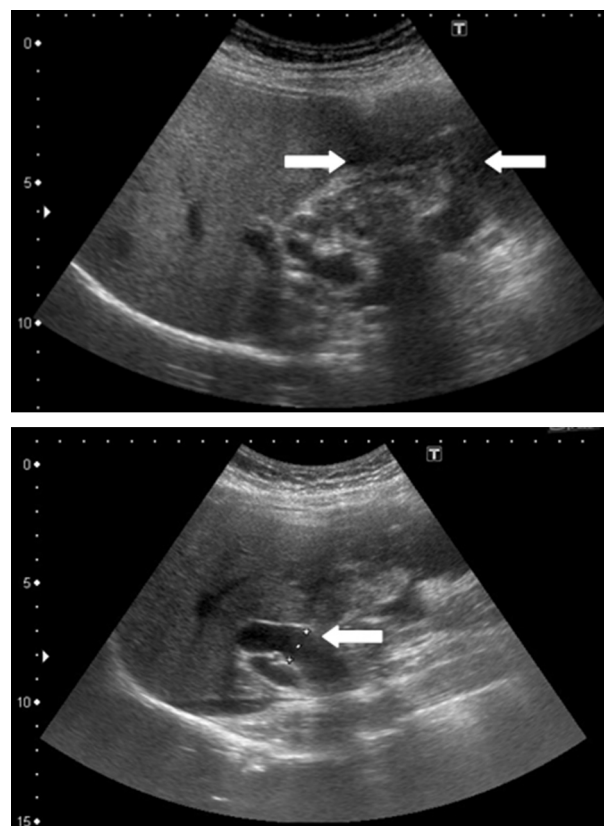


Figure 1 Abdominal ultrasonography showing one mixed echogenic tumor approximately 9.5 cm from the head of the gallbladder sac. The common bile duct was dilated to 1.4 cm.

prognosis of gallbladder sarcomatoid carcinoma is poor even after curative resection and there is as yet no known effective adjuvant therapy.

The risk factors for gallbladder sarcomatoid carcinoma are not clear. Risk factors for gallbladder carcinoma include adenomyomatosis, anomalous union of the pancreaticobiliary ductal system, cholelithiasis, chronic *Salmonella typhi* infection, a first-degree relative with gallbladder cancer, and exposure to carcinogens such as methylcholanthrene, *o*-aminoazotoluene and nitrosamines. Our patient did not have any of these risk factors. Patients with gallbladder sarcomatoid carcinoma often present with nonspecific symptoms and signs such as abdominal pain, nausea, anorexia, fever, jaundice, weight loss, or a palpable mass [1,6]. Varying CA 19-9 levels were noted among previous gallbladder sarcomatoid carcinoma cases [1,7]. Our patient was diagnosed at a late stage when she presented with abdominal pain and had an elevated CA 19-9 level that decreased after radical resection.

Because gallbladder sarcomatoid carcinoma is poorly understood, a definite staging system is not available. Most case series or reports are staged by the TNM classification of gallbladder carcinoma. The most important treatment is surgical resection. Our patient underwent a radical resection; adjuvant chemotherapy or radiotherapy were not used due to a lack of evidence supporting a therapeutic benefit [6,8]. The use of fluorouracil plus oxaliplatin for adjuvant therapy was previously documented in another

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