Simultaneous osteosarcoma and adenocarcinoma of the gallbladder: A rare case report and literature review

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Dear Editor,

Carcinosarcomas are rare malignant tumors that contain both epithelial and mesenchymal components (1) and usually develop in areas such as the head or neck and the respiratory tract; however, the occurrence of gallbladder (GB) carcinosarcomas is exceptionally rare (2). Herein we present a case of GB carcinosarcoma that contained osteosarcoma as a mesenchymal component and was treated using surgical resection.

A 70-year-old woman presented with a history of right upper abdominal discomfort for several months. Computed tomography (CT) scans revealed the presence of multiple gallstones with a thickened GB wall and calcified periportal lymph nodes, which were associated with GB cancer (Figure 1). Physical examination and laboratory examination revealed no abnormal findings. The serum level of tumor marker including carcinoembryonic antigen and carbohydrate antigen 19-9 were 3.08 ng/mL and 0.6 U/mL, respectively. The initial diagnosis was GB cancer and the differential diagnosis included porcelain GB and calcified calculus chronic cholecystitis. The patient underwent extended cholecystectomy, segmental resection, and anastomosis of the transverse colon because of direct invasion of the transverse colon by the tumor.

Histopathologic examination of the mass showed a predominantly epithelial component with an adenocarcinoma and a sarcoid component with osteosarcoma (Figure 2). The tumor invaded the transverse colon, and six metastatic lymph nodes were detected around the common hepatic artery and peripancreatic areas. Immunohistochemically, the adenocarcinoma showed positive staining for cytokeratin and carcinoembryonic antigen, and the osteosarcoma showed positive staining for vimentin (Figure 3). The patient was eventually diagnosed with a carcinosarcoma, which was classified as IVB (T3N2M0) per the 8th American Joint Committee on Cancer Tu-

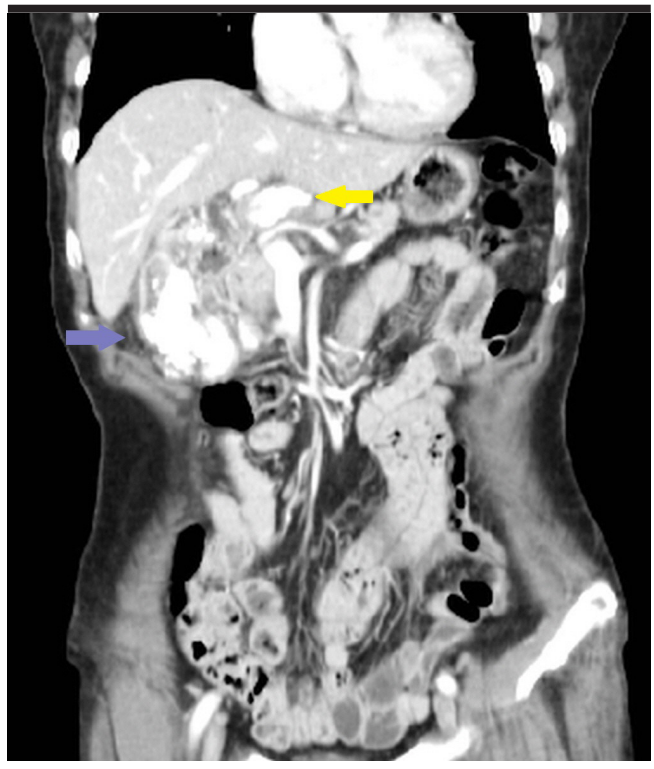

Figure 1. Contrast-enhanced abdominal computed tomography scan shows multiple gallstones with gallbladder wall thickening (purple arrow) and calcified periportal lymph nodes (yellow arrow), which is suggestive of gallbladder cancer

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mor-Node-Metastasis staging system. On day 13 following surgery, a follow-up CT scan showed the aggravation of enlarged lymph nodes around the para aortic areas and pancreatic head metastasis. She was discharged on day 36 following surgery. Two months later, a follow-up CT scan revealed the aggravation of metastasis. Her general

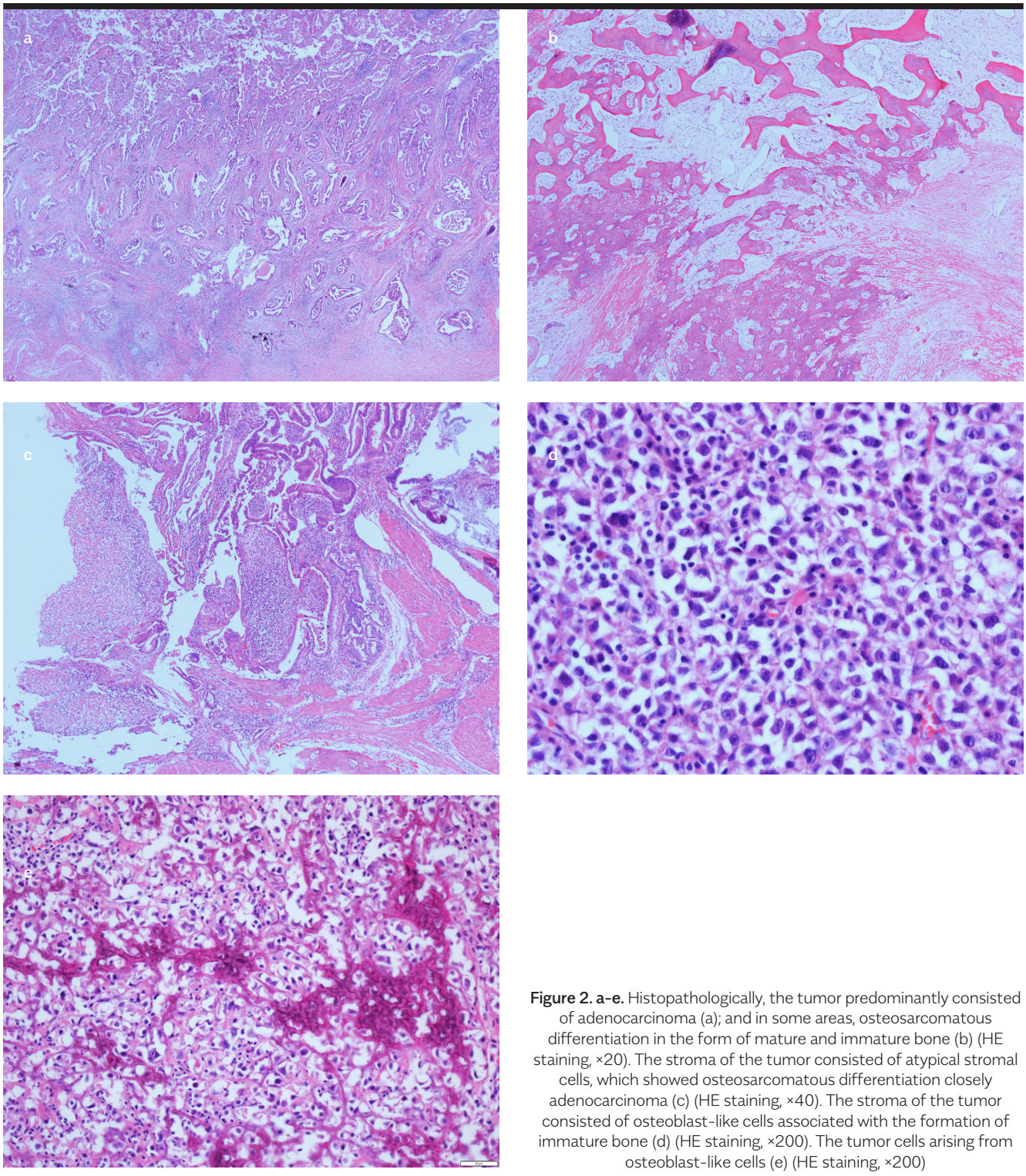

Figure 2. a-e. Histopathologically, the tumor predominantly consisted of adenocarcinoma (a); and in some areas, osteosarcomatous differentiation in the form of mature and immature bone (b) (HE staining, ×20). The stroma of the tumor consisted of atypical stromal cells, which showed osteosarcomatous differentiation closely adenocarcinoma (c) (HE staining, ×40). The stroma of the tumor consisted of osteoblast-like cells associated with the formation of immature bone (d) (HE staining, ×200). The tumor cells arising from osteoblast-like cells (e) (HE staining, ×200)

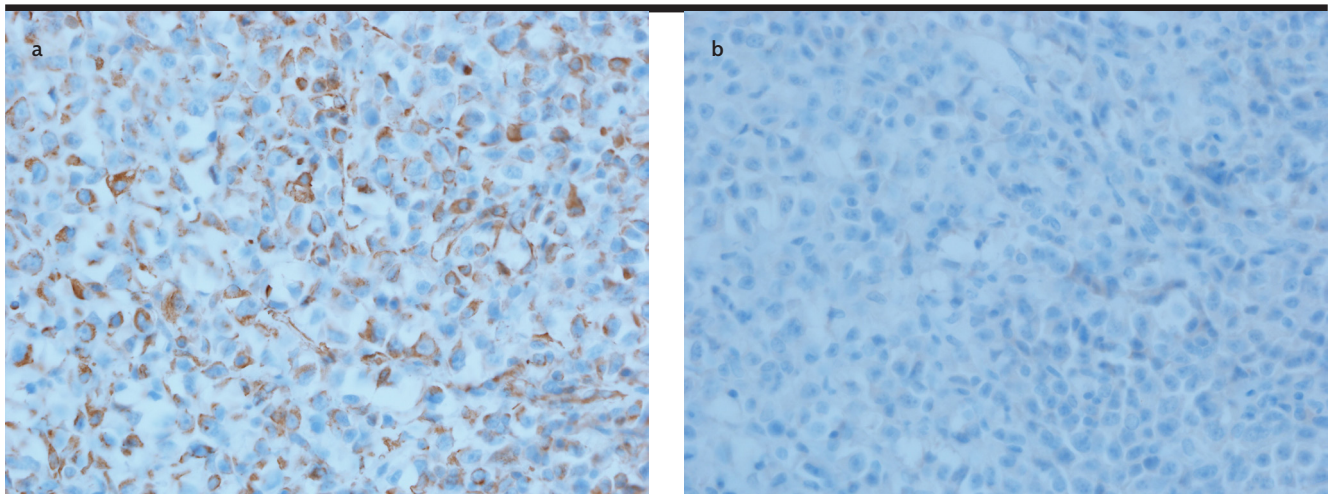

Figure 3. a, b. Osteosarcomatous area showed positive staining for vimentin (a); and negative staining for cytokeratin (b) (×400)

status and metastasis worsened, and she died 4 months following the surgery.

Carcinosarcomas with osteosarcoma of the GB are extremely rare. Carcinosarcomas are characterized by both epithelial and mesenchymal components within the same tissues. Microscopically, the diagnosis of carcinosarcoma requires the presence of both epithelial and mesenchymal components in the same tissue. Immunohistochemically, the epithelial component is positive for epithelial markers such as epithelial membrane antigen and cytokeratin, and the mesenchymal component is positive for mesenchymal markers such as actin, desmin, and vimentin.

The clinical presentation of GB carcinosarcoma is similar to that of GB carcinoma and includes symptoms of pain and the findings of a mass in the right upper abdomen, weight loss, general fatigue, and jaundice. The preoperative imagining studies such as ultrasonography, CT, MRI, and abdominal angiography could not differentiate GB carcinosarcoma from GB carcinoma. Therefore, the preoperative diagnosis of GB carcinosarcoma is challenging (1).

Surgical resection is the only curative option for GB carcinosarcomas and GB adenocarcinomas. Patients need to undergo extended cholecystectomy and en bloc lymph node resections because carcinosarcomas can recur as systemic metastases to the liver and lymph nodes. Effective adjuvant treatments are not known (1,3-5). The prognoses of GB carcinosarcomas are poor. The mean survival duration of patients with GB carcinosarcomas is only a few months (1,2). In a patient with a stage IV N2 carcinosarcoma, multiple metastatic lymph nodes were found 6 days following surgery, and the patient died 4 months later. However, some patients who have early-stage disease may achieve long-term survival (1).

In conclusion, we report a case of gallbladder carcinosarcoma with an osteoid component, which was treated by surgical resection. The preoperative diagnosis is very difficult because of the absence of specific radiological findings or tumor markers. Gallbladder carcinosarcomas have an aggressive nature and poor prognoses. Radical resection is still the mainstay of therapy for this disease in patients who are eligible for surgery.

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