



Acute pancreatitis due to pancreatic metastasis of osteosarcoma: a report of two cases

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Abstract: Metastasis of osteosarcoma to the pancreas is quite rare, and there have been no reports of metastasis-induced acute pancreatitis (MIAP) due to pancreatic metastasis of osteosarcoma. Here, we present two cases of MIAP due to pancreatic metastasis of osteosarcoma. Case 1: a 47-year-old woman was referred to our hospital due to osteosarcoma at left femur. She felt back pain 2 years postoperatively. An abdominal CT showed a 7 cm mass in the pancreatic head. Considering that the acute pancreatitis due to pancreatic metastasis, palliative radiotherapy was performed. Although abdominal pain was improved, she had died of multiple metastasis after 2 months. Case 2: a 42-year-old woman was referred to our hospital due to osteosarcoma at left femur. She felt abdominal and back pain 6 months after initial treatment. CT scans showed a large mass in the pancreatic tail. The mass increased rapidly over a short duration and complicated the acute pancreatitis. Palliative radiotherapy (30 Gy/10 fractions) was performed. After 1 month, the patient died of lung metastases. In previous reports, all patients with pancreatic metastases from osteosarcoma developed metastases at other sites, such as the lungs and bone. Therefore, we suggest that follow-up examinations at the abdominal lesion may be necessary for patients with metastatic disease.

Keywords: Osteosarcoma; pancreas; metastasis; acute pancreatitis

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Introduction

Pancreatic metastasis is frequently reported in lung cancer, gastrointestinal malignancies, breast cancer, renal cell carcinoma, melanoma, lymphoma, and soft tissue sarcoma. Osteosarcoma is the most common malignant bone tumor in children and adolescents. The most common sites of metastasis are the lungs (1-3). Metastatic pancreatic tumors are uncommon, with an incidence of 1.6–11% in autopsy studies (4). Acute pancreatitis due to pancreatic metastasis has been reported as metastasis-induced acute pancreatitis (MIAP) (5-8). The mechanism of MIAP may be pancreatic ductal obstruction due to tumor growth, impaired blood flow by the direct tumor invasion, or/and enlarged peripancreatic lymph nodes (7). There have been reports of acute pancreatitis due to pancreatic metastasis of various carcinomas such as small cell lung cancer, hepatocellular carcinoma, and renal cell carcinoma (5-8). Metastasis of

osteosarcoma to the pancreas is quite rare, and there have been no reports of MIAP due to pancreatic metastasis of osteosarcoma. Here, we present two cases of MIAP due to pancreatic metastasis of osteosarcoma. We obtained written informed consent from patients for present report when they received surgery.

Case presentation

Case 1

A 47-year-old woman was referred to our hospital due to left femur pain. A diagnosis of osteosarcoma of the distal femur was made with open biopsy. Wide tumor resection and reconstruction with endoprosthesis were performed. The pathological findings at surgery showed high-grade osteosarcoma, and she was treated with adjuvant chemotherapy using doxorubicin and ifosfamide

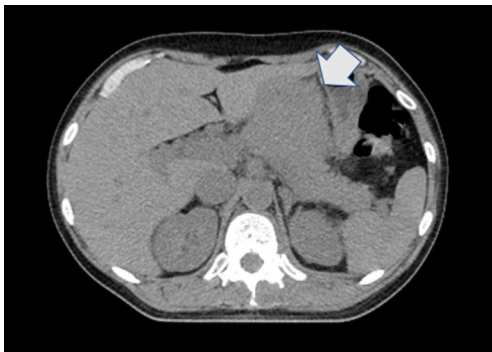


Figure 1 The CT showed pancreatic metastasis (arrow) in case 1.

postoperatively. However, 1 year and 4 months after initial treatment, the patient experienced lower back pain, and bone metastases were found in the twelfth thoracic vertebral body. A chest CT scan showed that she also had multiple lung metastases. Chemotherapy using gemcitabine and docetaxel was performed, followed by carbon ion radiotherapy for the twelfth thoracic vertebral body. However, the patient still felt back pain 2 years postoperatively. An abdominal CT showed a 7 cm mass in the pancreatic head (*Figure 1*). Laboratory examination showed increased level of amylase (340 U/L). Considering that the acute pancreatitis due to pancreatic metastasis, palliative radiotherapy (30 Gy/10 fractions) was performed. Although abdominal pain was improved, she had died of multiple metastasis after 2 months.

Case 2

A 42-year-old woman presented with left knee pain. After X-ray and MRI were performed in the clinic, she was referred to our hospital. A diagnosis of high-grade osteosarcoma was confirmed by open biopsy at the distal part of the femur. She was treated with three cycles of neo-adjuvant chemotherapy using doxorubicin and ifosfamide, followed by wide tumor resection and distal femur replacement. Although postoperative adjuvant chemotherapy was administered, she developed lung metastases 3 months after the initial treatment. Radiofrequency ablation was performed for the multiple lung metastases. However, she felt abdominal and back pain 6 months after initial treatment. CT scans showed a large mass in the pancreatic tail (*Figure 2*). The mass increased rapidly over a short duration and complicated the acute pancreatitis. The patient's levels of amylase were also elevated (200 U/L). Palliative radiotherapy (30 Gy/10

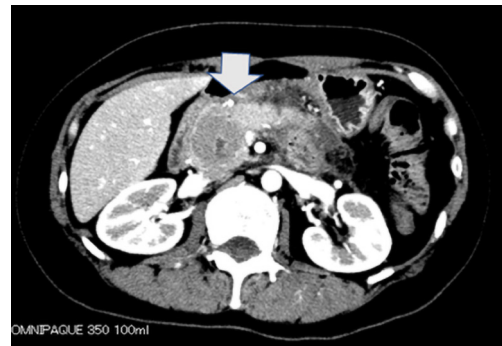


Figure 2 The CT showed pancreatic metastasis (arrow) in case 2.

fractions) was performed. After 1 month, the patient died of lung metastases.

Discussion

We presented two case reports of acute pancreatitis due to pancreatic metastasis in patients with osteosarcoma. There are some case reports concerning pancreatic metastasis of osteosarcoma (3,9-15) (*Table 1*). Ogose *et al.* reported 223 patients with osteosarcoma between 1975 and 1998, with two patients (0.8%) developing pancreatic metastases (16). Yoon *et al.* reported 53 patients with pancreatic metastasis between 1997–2009, with only one patient having primary osteosarcoma (17).

Metastases to the pancreas have various symptoms, such as jaundice, abdominal and back pain, vomiting, and nausea. However, some patients are asymptomatic. Metastases are found during regular follow-up examinations for their primary disease (3,11-13). In the present cases, the patients had severe abdominal pain, elevated levels of amylase, and pancreatic metastases combined with acute pancreatitis as diagnosed with CT scans.

The prognosis after pancreatic metastasis is generally poor, because patients will in most cases develop multiple metastases. In previous reports, data related to patient outcomes was limited due to a quite short follow-up. Although there is no evidence that resection for pancreatic metastasis in osteosarcoma is superior to an alternative, surgical management was reported to be effective in patients with renal cell carcinoma (17). If metastasis can be controlled in patients with osteosarcoma, surgery and/or chemotherapy may be indicated.

Acute pancreatitis due to pancreatic metastasis has been reported as MIAP (18). Most cases of MIAP were diagnosed clinically, because tissue diagnosis may be difficult in MIAP

Table 1 The list of the osteosarcoma patients with pancreatic metastasis

Ref	Age	Sex	Symptoms	Other metastases	Biopsy	Metastasectomy	Outcome
(3)	44	F	None	Lung	Yes	Yes	Alive, 14 months
(9)	25	F	Melaena	Lung	No	Yes	Alive, 11 months
(10)	13	F	Vomiting	Lung	No	Yes	Died, 18 months
(11)	53	M	none	Lung	Yes	Yes	Alive, 10 months
(12)	33	M	none	Lung	Yes	No	Alive, 5 months
(13)	28	M	None	Brain	No	No	Died, 8 months
(13)	66	F	Jaundice	Lung, liver	No	Yes	Unknown
(14)	14	M	Nausea	Bone	No	No	Died, 2 weeks
(15)	18	M	Abd. pain	Lung	Yes	Yes	Alive, 7 months
Our case	47	F	Back pain	Lung, bone	No	No	Died, 2 months
Our case	42	F	–	Lung	No	No	Died, 1 months

Ref, reference; M, male; F, female.

due to poor performance status of the patients (19). MIAP is quite rare, with Stewart *et al.* reporting only one patient diagnosed with MIAP out of 802 lung cancer patients. They also suggested that treatment for pancreatitis should be first considered, and once the pancreatitis is alleviated, treatment for the metastatic disease can be performed (19). In the present cases, palliative radiotherapy was performed, because the patients developed multiple metastases and no further treatment was planned. In previous reports, all patients with pancreatic metastases developed metastases at other sites, such as the lungs and bone. Therefore, we suggest that follow-up examinations at the abdominal lesion may be necessary for patients with metastatic disease.

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Footnote

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Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). We obtained written informed consent from patients for present report when they received surgery.

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