Clinical Case Report

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Long-term survival after sporadic and delayed metastases of conventional osteosarcoma

A case report

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Abstract

Histologically conventional osteosarcoma, once metastasized to the lung, generally causes a rapid and fatal outcome. Osteosarcoma metastasis to the gastrointestinal tract is extremely rare.

We report herein a case of osteoblastic osteosarcoma with exceptionally unique features: sporadic lung metastases and delayed metastases to the stomach and the jejunum with long-term survival. She received multiple operations and chemotherapies, but consequently died of peritoneal dissemination. A review of the literature on osteosarcoma metastasis to the gastrointestinal tract is presented.

This patient was very unusual in terms of a long-term survival and metastatic sites, suggesting the importance of vigilance and thorough follow-up for patients with conventional osteosarcoma.

Abbreviations: CT = computed tomography, MRI = magnetic resonance imaging.

Keywords: case report, gastrointestinal tract, jejunum, metastasis, osteosarcoma, stomach

1. Introduction

Osteosarcoma is the most common type of bone tumor in children and the lung is the most frequent site for metastatic osteosarcoma. Histologically conventional osteosarcoma, once metastasized to the lung, generally causes a rapid and fatal outcome. We report herein a case of osteoblastic osteosarcoma with exceptionally unique features: sporadic lung metastases and delayed metastases to unusual sites with long-term survival.

2. Case presentation

An 18-year-old female patient presented with a history of pain in the left knee in August 2002. Magnetic resonance imaging and open biopsy revealed osteoblastic osteosarcoma in the proximal tibia (Fig. 1). She was treated with wide resection of the tumor and perioperative chemotherapy, including cysplatin, doxorubicin, and methotrexate. However, 2 years later pulmonary metastasis was detected and treated with surgical resection and ifosfamide chemotherapy in October 2004. Then, she was treated for 5 more events of pulmonary metastasis by repeated operations and adjuvant conventional chemotherapies for

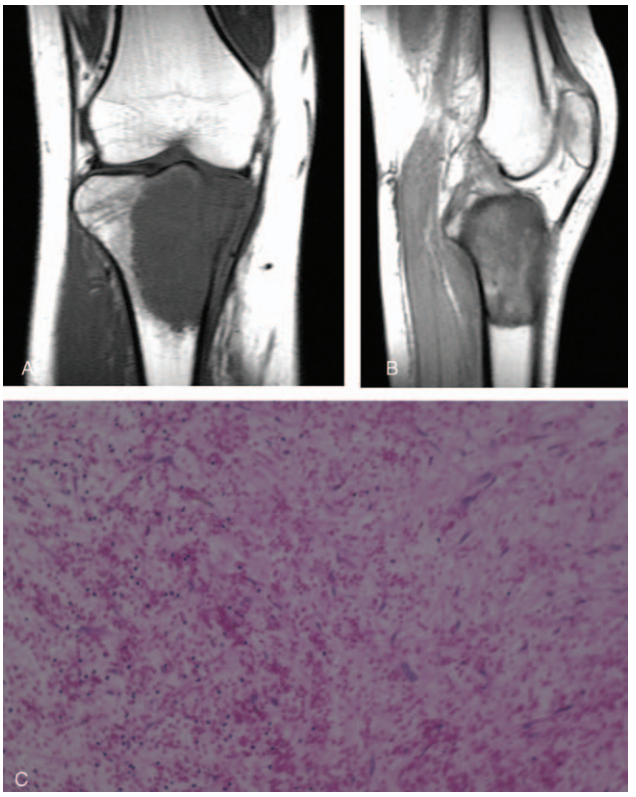

Figure 1. Anteroposterior and lateral MRI of the left knee reveal a bone tumor of the proximal tibia (A and B). A microphotograph shows pleomorphic spindle-shaped atypical cells with osteoid (C). MRI = magnetic resonance imaging.

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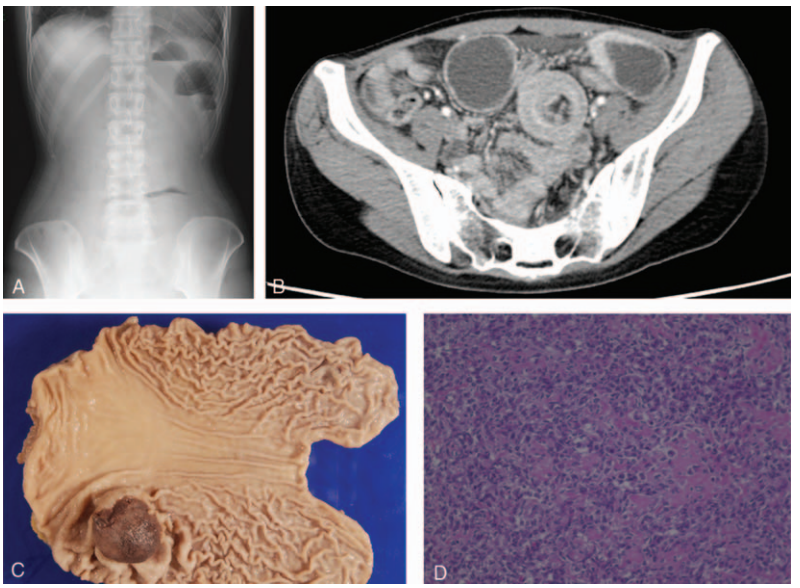

Figure 2. Erect abdominal radiography shows small bowel loops with stepladder pattern (A). Abdominal CT demonstrates jejunal intussusception caused by a tumor (B). The size of the jejunum tumor was 2 cm × 2 cm ×2 cm (C). A microphotograph proves the existence of metastatic osteosarcoma cells (D). CT = computed tomography.

sarcoma, including ifosfamide, etoposide, and gemcitabine (July 2005, May 2007, January 2008, June 2008, January 2012).

In November 2011, she consulted a general hospital complaining of tarry stools. A gastrointestinal endoscopic biopsy revealed osteosarcoma metastasis to the stomach. Subsequently, total gastrectomy was performed. In September 2012, she was referred to our emergency department with abdominal pain and vomiting. Abdominal computed tomography scans showed jejunal intussusception caused by a tumor. She then underwent laparoscopic tumor resection. An intraluminal metastatic osteosarcoma was found at the point of intussusceptions (Fig. 2). She subsequently received adjuvant chemotherapy including gemcitabine and docetaxel. Unfortunately, she died of peritoneal dissemination in May 2015. Her family did not accept our request for an autopsy.

The patient written informed consent was waived due to the retrospective nature of the presented case. Patient information was anonymized and deidentified.

3. Discussion

The most common site for metastatic osteosarcoma is the lung, occurring in more than 90% of cases. Other sites of metastases include the bones, pleura, brain, and intraabdominal solid viscera.[1] Osteosarcoma metastasis to the gastrointestinal tract is extremely rare. To our knowledge, there are only 20 previously documented cases. Of these, 5 were to the stomach,[2–6] 3 were to the duodenum,[7–9] 8 were to the jejunum,[10–17] and 3 were to the ilium.[18–21] The metastasis localized in the stomach presented as anemia, hematemesis, and tarry stools. In 5 cases, patients underwent a resection of the metastatic masses endoscopically or using a laparotomy or laparoscopy to determine each tumor's size and localization. The metastasis in the jejunum presented as abdominal pain and vomiting, and in 7 of 9 cases intussusception occurred. Controversy exists regarding whether intussusception can be treated conservatively. Chandramohan et al[15] reported that they could not conservatively cure their patient of intussusception caused by osteosarcoma metastasis to the jejunum, so they concluded that surgical management should be considered because of the risk of gangrene and perforation.

Recent aggressive therapy and increased long-term survival have brought about a change in the natural history of osteosarcoma. This, in turn, might have led to a recent increase in reports on osteosarcoma metastasis to the gastrointestinal tract. Giuliano et al[22] addressed the fact that advances in therapy such as multi target chemotherapy have altered the pattern of recurrences and metastasis in patients treated for osteosarcoma. Bacci et al reported on 789 osteosarcoma patients treated at a single institution. Of those 789 cases, 313 patients experienced recurrence. Of these 62 patients went into remission and 246 patients died after repeated remissions and relapses. The mean survival of the latter patients was about 3 years from the last recurrence, with the maximum relapse or recurrence rate being 7.[1] The patient described here survived 11 years after the first recurrence and experienced 8 relapses. This patient was very unusual in terms of a long-term survival and metastatic sites, suggesting the importance of vigilance and thorough follow-up for patients with conventional osteosarcoma.

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