

Intestinal Metastasis of Osteosarcoma Presenting with Intussusception

- A Case Report -

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Metastatic osteosarcoma most commonly affects the lungs and other bones. Intestinal intussusception caused by metastatic osteosarcoma is very rare. We report a case of metastatic osteosarcoma of the intestine in a 39-year-old female. She underwent surgical resection of the left femur due to osteosarcoma and received additional chemotherapy 3 years ago. Pulmonary metastasis was found two years later and the patient complained of abdominal pain, nausea and vomiting after 8 months following excision of the lung nodules. Abdominal computed tomography revealed intussusception with a suspected polypoid mass in the distal portion of the jejunum. The histologic findings of the resected bowel were those of osteosarcoma. This is the first case of documented intestinal metastasis of osteosarcoma in Korea. It is suggested that the tumor metastasis to the small intestine should be considered in patients with previous osteosarcoma, when the patient presents with acute abdominal symptoms and intussusception.

Key Words : Osteosarcoma-Neoplasm Metastasis-Intussusception

The lung is the most common metastatic site of osteosarcoma, and approximately 90% of fatalities in patients with osteosarcoma is due to pulmonary metastases.¹ However, gastrointestinal metastases is extraordinarily rare. Recently the development of therapeutic multimodalities increased the incidence of extrapulmonary metastasis of osteosarcoma along with the prolongation of event-free survival of the patients.¹⁻³ We report on a patient with osteosarcoma on the left femur who subsequently showed intussusception, and discuss its clinicopathologic significance.

CASE REPORT

A known 39-year-old female osteosarcoma patient presented with abdominal pain, nausea, and vomiting. Three years ago, she felt pain and swelling of the left knee and the diagnosis of osteosarcoma was made from excisional biopsy of the left femur.

There was no evidence of metastasis at that time and the patient received two cycles of neoadjuvant chemotherapy with high-dose methotrexate. Subsequently, surgical excision and tumor prosthesis was performed. Microscopic examination of the resected bone showed chondroblastic osteosarcoma with tumor necrosis less than 30%. After the surgery, she received four cycles of high-dose methotrexate and one cycle of adriamycin. Two years later, follow-up computed tomography and bone scans revealed suspicious tumor thrombus in the left pulmonary artery, and four metastatic nodules in the left lower and right upper lobes of the lung. She received four more cycles of high-dose methotrexate after excision of the metastatic nodules. At 10 months after resection of the metastatic pulmonary nodules, the patient suffered from abdominal pain, nausea and vomiting. Abdominal computed tomogram (CT) scan disclosed intussusception with a suspected polypoid mass in the distal portion of the jejunum (Fig. 1). Segmental resection of the jejunum was performed.

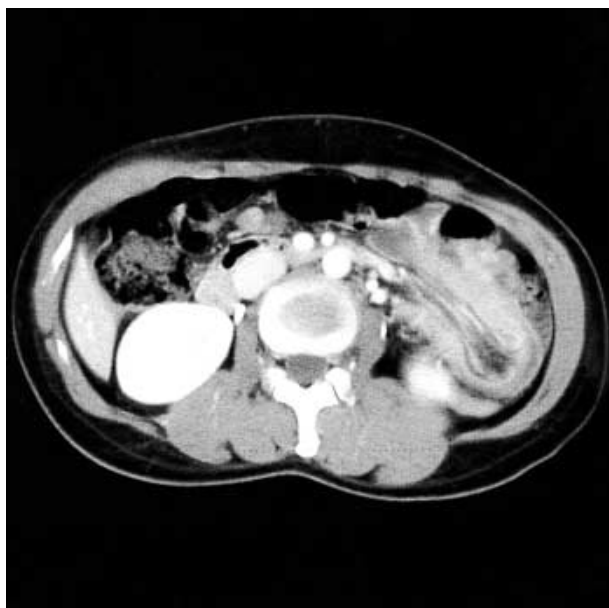


Fig. 1. Abdominal computed tomographic scan reveals jejunal intussusception in the left abdomen with marked swelling of the wall.



Fig. 2. An ulcerated and polypoid tumor mass, measuring $3 \times 3 \times 3$ cm, is noted in the distal jejunum.

Gross examination of the bowel revealed an ulcerated and polypoid mass, measuring $3 \times 3 \times 3$ cm (Fig. 2). The cut surface of the lesion was whitish and solid, and involved the full thickness of the jejunum without necrosis or calcification. Microscopically, the tumor infiltrated through the entire wall of jejunum and most of the crypts were destroyed (Fig. 3A). The tumor was composed of large pleomorphic, round to

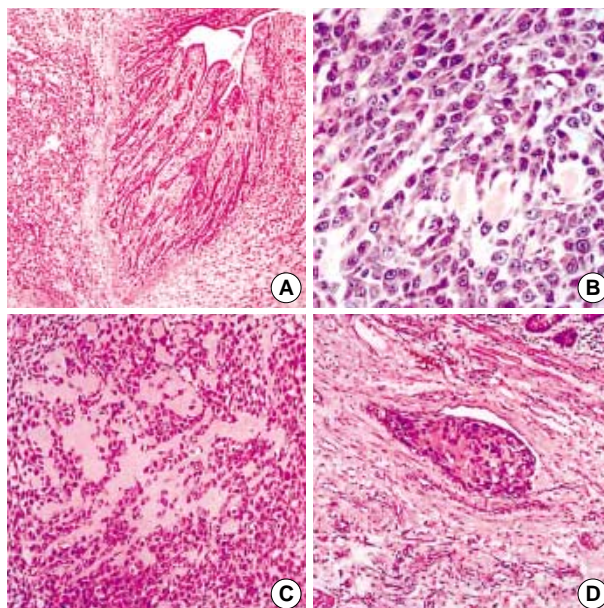


Fig. 3. (A) Polypoid tumor mass involves the entire wall of jejunum. (B) The tumor is mainly composed of large, irregular shaped cells with prominent nucleoli and high mitotic activity. (C) Osteoid is noted around tumor cells. (D) Lymphatic involvement by osteosarcoma is noted in the submucosa apart from the main tumor masses.

polygonal cells with plump eosinophilic cytoplasm. The eccentrically located nuclei were irregular, bizarre, and hyperchromatic with coarse chromatin and occasional prominent nucleoli. Mitotic figures were brisk (more than 20/10 high power field) and atypical forms were frequently noted (Fig. 3B). Fine eosinophilic lace-like osteoid and chondroid matrix surrounding the tumor cells were found (Fig. 3B, C). In the submucosa, vascular invasion and endolymphatic tumor emboli were frequently found (Fig. 3D).

DISCUSSION

At the time of the initial diagnosis, osteosarcoma is usually localized and distant metastasis is identified in only 10% to 20% of cases.⁴ As previously reported in the literature, most metastases are found in the lung and other sites of bones are the second most frequent site. Although extrapulmonary metastases to the brain, bones, pleura and heart can occur, these have been usually found in patients with preterminal diffuse disease or during autopsy.⁵ However, advances in therapeutic multimodalities including neoadjuvant and adjuvant chemotherapy improved patients' event-free survival resulting in a change of the pattern of metastases.^{2,3} The main route of metastatic osteo-

sarcoma is the hematogenous spread, however lymphatic metastases accounts for about 10% in large tumors.⁶ Intestinal metastasis can occur by lymphatic spread or hematogenous invasion. In our case, we observed both vascular invasion and frequent endolymphatic tumor emboli.

Intussusception is more common in pediatric cases but rare in adults, accounting for 1% to 5% of all causes of intestinal obstruction. The cause of intussusception differs between pediatric and adult populations. In the pediatric population, a precipitating lesion is found only in 10%, whereas the underlying cause is present in 90% of the adult population. Eisen LK *et al.*⁷ retrospectively renewed the cause of intussusception in 27 adult patients; 4 of 5 large bowel lesions were primary adenocarcinomas. In contrast, 8 of 22 small bowel lesions were malignant tumors and all small bowel cancers were metastatic disease. In our case, the metastatic tumor involve the jejunum. Metastatic tumors presented as intussusception include melanoma, liposarcoma, renal cell carcinoma, malignant fibrous histiocytoma, lung cancer, malignant lymphoma, and germ cell tumor,⁸⁻¹³ but metastatic osteosarcoma is very rare as a cause of intussusception.

In a literature survey, we found only 6 cases of intestinal metastatic osteosarcoma (one in the duodenum and 5 in the jejunum).¹⁴⁻¹⁹ Among them, 5 cases of metastatic osteosarcoma presented with intussusception, and the other was found during endoscopic examination. In our case, the metastasis was localized in the jejunum and clinical manifestation was acute abdominal symptoms. Mesenteric and peritoneal metastases have not been observed. Interestingly, all patients of intestinal osteosarcoma received adjuvant and neoadjuvant chemotherapy for primary osteosarcoma. These findings may support the changing pattern (intestinal metastasis) of metastatic osteosarcoma induced by chemotherapy.

In conclusion, the findings of our case suggest that the intussusception due to intestinal metastasis should be considered in patients with previous osteosarcoma in a clinical manifestation of acute abdominal symptoms. We also considered metastatic causes when we come into contact with intussusception of the small intestine in adult patients.

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