Disseminated metastatic disease of osteosarcoma of the femur in the abdomen: Unusual metastatic pattern on Tc-99m MDP bone scan

Ozgur Karacalioglu,* Seyfettin Ilgan,* Okan Kuzhan,** Ozdes Emer* and Mehmet Ozguven*

Departments of *Nuclear Medicine and **Medical Oncology, Gulhane Military Medical Academy and School of Medicine, Ankara, Turkey

A 25-year-old patient with osteosarcoma of the right distal femur underwent a bone scintigraphy with Tc-99m methylene diphosphonate (MDP). Whole-body bone scan revealed extensive metastatic disease in the abdominal region. Abdominal computerized tomography confirmed the presence of ascites and calcified masses on the greater omentum and peritoneal surfaces. Here we describe a case of unusual metastatic pattern of an osteosarcoma showing extensive intraabdominal metastases without prominent lung involvement after intensive chemotherapy.

Key words: Tc-99m MDP, osteosarcoma, metastasis, abdomen

INTRODUCTION

OSTEOSARCOMA is the most common primary bone tumor in children and young adults. 1 It is thought to arise from primitive mesenchymal bone-forming cells, and its histological hallmark is the production of malignant osteoid. A number of variants of osteosarcoma exist, including conventional types (osteoblastic, chondroblastic, and fibroblastic), telangiectatic, multifocal, parosteal, and periosteal. Not all osteosarcomas arise in a solitary fashion, as multiple sites may become apparent within a period of about 6 months (synchronous osteosarcoma), or multiple sites may be noted over a period longer than 6 months (metachronous osteosarcoma). Such multifocal osteosarcoma is decidedly rare, but when it occurs, it tends to be in patients younger than 10 years.² Over 90% of tumors are located in the metaphysis; the most common sites are the bones around the knee which account for 80% of cases. The mainstay of therapy is surgical removal of the malignant lesion. Most often, limb-sparing (limb-preserving) procedures can be used to treat patients with this disease and, thus, preserve function. Chemotherapy also is required to treat micrometastatic disease, which is

Received January 15, 2006, revision accepted April 24, 2006. For reprint contact: Ozgur Karacalioglu, M.D., Gulhane Military Medical Academy and School of Medicine, Department of Nuclear Medicine, 06018-Etlik, Ankara, TURKEY.

E-mail: aokaracali@yahoo.com

present but often not detectable in most patients (about 80%) at the time of diagnosis.

Bone scan is a cost-effective and sensitive method of choice for osseous metastases as well as metastases in soft tissues of an osteosarcoma.^{3,4} It can also help to differentiate nonspecific calcified pulmonary or other soft tissue lesions such as calcified granulomas from osseous metastases. Furthermore, the bone scan can allow the detection of the rare multifocal variant of osteosarcoma or unusual metastatic patterns of this tumor as presented in this case report.

CASE REPORT

A localized osteosarcoma of the right distal femur was diagnosed in a 25-year-old woman (Fig. 1a and Fig. 1b). The patient received high dose chemotherapy. During the treatment, the primary extremity tumor was removed and an endoprosthesis was placed. She received autologous peripheral blood stem cell transfusion 3 months after the initial diagnosis. Although the engraftment was successful with no serious side effects, she developed severe bone pain, and first relapse was detected in the bones 1 year later (Fig. 2a). Intensive chemotherapy including methotrexate was repeated. After treatment, Tc-99m MDP bone scan was required for control purpose and whole-body bone scan 4 hours after the injection of 20 mCi (740 MBq) Tc-99m MDP revealed extensive metastatic disease in the abdominal region and in bones of the patient (Fig. 2b).

Vol. 20, No. 6, 2006 Case Report 437

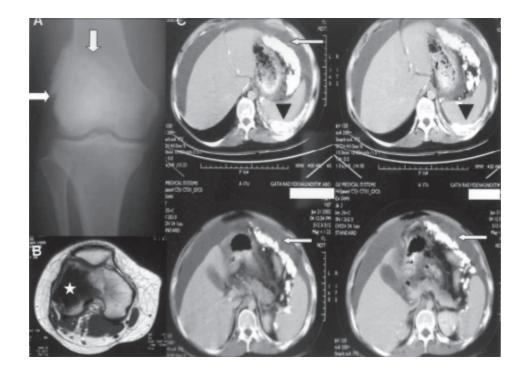


Fig. 1 The x-ray of the primary tumor demonstrates the calcification within the mass (arrows) (A), and the MRI scan confirms that this osteosarcoma arises from the right distal femur (star) (B). Abdominal computerized tomography showing the presence of metastatic deposits in peritoneal cavity (arrowheads) and along the surface of the greater omentum (arrows) (C). Note the diffuse nature and punctuated pattern of the lesions.

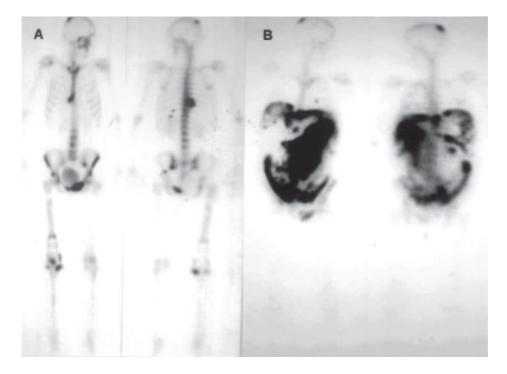


Fig. 2 Previous anterior and posterior whole-body bone scan of the patient shows multiple metastatic foci in her skeleton (A). The latest anterior and posterior whole-body bone scan of the patient demonstrates several regions of focal and diffuse intense uptake of varying size and intensity in the abdominal region. Interestingly, there is only one focal activity uptake in the left hemithorax (B).

The intraabdominal lesions demonstrated abnormal avidity for Tc-99m MDP on bone scintigraphy indicating extensive metastases. Surprisingly, the lungs were almost spared. Abdominal computed tomography and ultrasonography disclosed ascites and multiple calcified masses on the peritoneum and on the greater omentum (Fig. 1c). Due to disease progression, her condition deteriorated with severe metastatic disease and the patient died of progressive disease 3 years after the initial diagnosis.

DISCUSSION

Osteosarcoma is a deadly form of musculoskeletal cancer that most commonly causes patients to die of pulmonary metastatic disease. At diagnosis, 80% of patients have localized disease, while distant metastases can be identified in only 10%.⁵ Involvement of local or regional lymph nodes is unusual. The most frequent sites of metastases are the lungs and bones.6 Lung metastases for osteosarcoma have been occasionally associated with lesions in bone, brain, chest wall, lymph node, and skin. Despite the dramatic improvement in the treatment of patients with osteosarcoma, approximately 25 to 30% of these tumors recur after therapy. At the time of initial recurrence, pulmonary disease is the most common finding in patients treated with adjuvant chemotherapy.^{6,8,9} In addition, aggressive attempts at salvage after relapse can cause metastases in other extrapulmonary sites, such as the spine, brain, and heart. 10,11 Clinical presentation of recurrent extrapulmonary metastasis of osteosarcoma is rare. In a few cases, metastases to chest wall, epidural space, brain, skin, heart, kidney, liver, mesentery, small intestine pleura and ovary have been reported. 12-20 Peritoneal metastases are very rare in osteosarcoma. 10,21-24

The osteosarcoma originating from the femur preferred the peritoneal seeding instead of lung involvement in our case. Although, neither abdominal nor retroperitoneal disease at diagnosis or relapse was detected in a retrospective review of 279 patients with osteosarcoma, 23 the coexistence of metastatic osteosarcoma of lung with peritoneal spread has been reported rarely. 19,23 In another case report, extraskeletal retroperitoneal osteosarcoma causing extensive peritoneal and hepatic metastatic foci has been reported.²⁵ It was possible that chemotherapy or radiation therapy may have changed the metastatic pattern of osteosarcoma after prolonged survival.²⁶ In one case report, a patient with telangiectatic osteosarcoma of the proximal fibula while receiving adjuvant chemotherapy, developed pulmonary and, later, mesenteric metastases and it has been reported that this case represents an additional example of an alteration in the clinical pattern of metastases induced by chemotherapy.¹⁵

As the initial treatment for patients with osteosarcoma is intensified, the pattern of metastases may change and unusual sites of recurrence may become more prevalent. Direct seeding into peritoneum by metastatic osteosar-

coma might be rare, but nevertheless was responsible for the abdominal metastasis in this case, which illustrates an unusual course of metastatic osteosarcoma and the importance of frequent abdominal survey in such patients. Therefore, patients with osteosarcoma with signs or symptoms of intestinal obstruction or an acute abdomen should undergo prompt and careful investigations, including a bone scan, to exclude peritoneal metastatic disease. In these patients, bone scan can play an important role in follow-up and it can reveal unexpected metastatic foci of this tumor.

REFERENCES

- Vander Griend RA. Osteosarcoma and its variants. Orthop Clin North Am 1996; 27: 575–581.
- Weis LD. Common malignant bone tumors: osteosarcoma. In: Surgery for Bone and Soft-Tissue Tumors. Simon MA, Springfield D (eds), Philadelphia; Lippincott-Raven, 1998: 265–274.
- 3. Brown ML. Bone scintigraphy in benign and malignant tumors. *Radiol Clin North Am* 1993; 31: 731–738.
- Wang K, Allen L, Fung E, Chan CC, Chan JC, Griffith JF. Bone scintigraphy in common tumors with osteolytic components. *Clin Nucl Med* 2005; 30: 655–671.
- 5. Marina N, Prat C, Rao B. Improved prognosis of children with osteosarcoma metastatic to the lung(s) at the time of diagnosis. *Cancer* 1992; 70: 2722–2727.
- Jeffree GM, Price HG, Sisson HA. The metastatic patterns of osteosarcoma. Br J Cancer 1975; 32: 87–107.
- Tabone MD, Kalifa C, Rodary C. Osteosarcoma recurrences in pediatric patients previously treated with intensive chemotherapy. *J Clin Oncol* 1994; 2: 2614–2620.
- 8. Jaffe N, Smith E, Abelson HT, Frei E 3rd. Osteogenic sarcoma: alterations in the pattern of pulmonary metastases with adjuvant chemotherapy. *J Clin Oncol* 1983; 1: 252–254.
- 9. Pratt C, Shanks E, Hustu O, Rivera G, Smith J, Kumar AP. Adjuvant multiple drug chemotherapy for osteosarcoma of the extremity. *Cancer* 1977; 39: 51–57.
- Bacci G, Avella M, Picci P. Metastatic patterns in osteosarcoma. *Tumori* 1988; 74: 421–427.
- Danzinger J, Wallace S, Handel SF, deSantos LA. Metastatic osteogenic sarcoma to the brain. *Cancer* 1979; 43: 707–710.
- 12. Nouyrigat P, Berdah J, Roullet B, Olivier J. Osteosarcoma with calcified regional lymph nodes. *Pediatr Radiol* 1993; 23: 74–75.
- Daw NC, Kaste SC, Hill DA, Kun LE, Pratt CB. Metastatic osteosarcoma to the liver after treatment for synovial sarcoma: a case report. *Pediatr Hematol Oncol* 2001; 18: 123– 128.
- Lockhart SK, Coan JD, Jaffe N, Eftekhari F, David C, Shirkhoda A. Osteosarcoma metastatic to the kidney. *Clin Imaging* 1989; 13: 154–156.
- Takaue Y, Slopis JM, Anzai T, Robertson R, Jaffe N. Successful treatment of pulmonary and abdominal metastatic osteosarcoma. *Med Pediatr Oncol* 1985; 13: 126–128.
- 16. Panizo-Santos A, Sola I, Lozano M, de Alava E, Pardo J.

Vol. 20, No. 6, 2006 Case Report 439

- Metastatic osteosarcoma presenting as a small-bowel polyp. A case report and review of the literature. Arch Pathol Lab Med 2000; 124: 1682-1684.
- 17. Eltabbakh GH, Belinson JL, Biscotti CV. Osteosarcoma metastatic to the ovary: a case report and review of the literature. Int J Gynecol Pathol 1997; 6: 76-78.
- 18. Uribe-butero G, Russell W, Sutow W. Primary osteosarcoma of bone: a clinicopathological investigation of 243 cases, with necropsy studies in 54. Am J Clin Pathol 1977; 67: 427-435.
- 19. Lin JT, Yen CC, Wang WS, Chiou TJ, Liu JH, Wu HT, et al. Case report: Unusual peritoneal spreading by metastatic osteosarcoma of the tibia. Br J Radiol 2003; 76: 337-338.
- 20. Costa CM, de Camargo B, Bagietto R, Alcantra PS, Chojniak R, Sredni ST. Abdominal recurrence of osteogenic sarcoma: a case report. J Pediatr Hematol Oncol 1998; 20: 271-273.

- 21. Teates C, Brower A, Williamson B. Osteosarcoma extraosseous metastases demonstrated on bone scan and radiographs. Clin Nucl Med 1977; 2: 298-302.
- 22. Hough D. Multifocal osteosarcoma with extensive pleural metastatic disease. Austral Radiol 1992; 36: 147-149.
- 23. Westra A, Schrijvers D, Somville J, Van Schil P, Hubens G, Van Oosterom A. Late peritoneal metastasis in a patient with osteosarcoma. Ann Oncol 1998; 9: 907-911.
- 24. Spieth ME, Ansari AN, Forrester DM, Kimura RL, Assassa GS, Siegel ME. Osteosarcoma presenting as intestinal obstruction. J Nucl Med 1993; 34: 1990-1991.
- 25. Maredia R, Ward DL, Moreno AJ, Carpenter AL. Extensive extraskeletal osteosarcoma in the liver and abdomen as demonstrated by bone scintigraphy. Clin Nucl Med 1997; 22: 717-718.
- 26. Giuliano AE, Feig S, Eilber FR. Changing metastatic pattern of osteosarcoma. Cancer 1984; 54: 2160-2164.