Case Report
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Osteopoikilosis: A Sign Mimicking Skeletal Metastases in a Cancer Patient

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Abstract

Osteopoikilosis is a rare benign osteosclerotic bone disorder that may be misdiagnosed as skeletal metastases. Here we describe a case of coincidental breast cancer and osteopoikilosis mimicking skeletal metastases. A 41-year-old woman underwent right modified radical mastectomy in April 2007. Twenty-eight months after initial treatment, the patient complained of bilateral knee and foot pain. Plain X-rays of the feet and knees showed multiple well-defined osteosclerotic lesions. According to the radiographic appearance, the most likely differential diagnoses included skeletal metastases from breast cancer and osteopoikilosis. A whole-body bone scintigraphy showed no increase in uptake by the sclerotic lesions, and serum lactic dehydrogenase, carcinoembryonic antigen, alkaline phosphatase and cancer antigen 15-3 were not elevated. We therefore diagnosed the patient's skeletal lesions as osteopoikilosis. This case and our literature review suggest that the radiographic appearance of osteopoikilosis may mimic or mask skeletal metastases, potentially leading to misdiagnosis in patients with cancer.

Keywords: Osteopoikilosis; Osteosclerotic; Skeletal metastases; Breast cancer.

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Introduction

Osteopoikilosis is a rare benign, autosomal-dominant genetic bone disorder typically characterized by widespread, numerous, small, well-defined oval or circular radiodensities throughout the skeleton. These bone lesions are usually asymptomatic and are often discovered incidentally on radiography. The sites involved most frequently are the epiphyses and metaphyses of long tubular bones, and the carpal, tarsal and pelvic

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bones; however, the skull, ribs and vertebral bodies are usually spared. Typically, in osteopoikilosis, bone scintigraphy is negative and biochemical parameters are within normal limits.[1]

The skeleton is the most frequent site of breast cancer metastases. The diagnosis of skeletal metastases in breast cancer is based on both radiological findings and clinical manifestations, supported by biochemical profiles. On conventional radiography, skeletal metastases of breast cancer may have osteolytic, osteoblastic or mixed appearances. Whole-body bone scintigraphy is the most useful method for detecting skeletal metastases in patients with breast cancer and evaluating the extent of metastatic disease. Bone scintigraphy is a particularly useful indicator of osteoblastic activity, although it is less useful in patients with purely osteolytic lesions.[2] Biochemical parameters such as carcinoembryonic antigen (CEA), alkaline phosphatase (AP), cancer antigen 15-3 (CA 15-3) and lactic dehydrogenase (LDH) are commonly altered in patients with skeletal metastases and are thus useful as complementary studies.[3]

The radiographic appearance of osteopoikilosis may be deceptive, leading to misdiagnoses in oncologic settings.[4] Here we describe a patient with coincidental breast cancer and osteopoikilosis that mimicked skeletal metastasis.

Case report

In April 2007, a 41-year-old woman presented with a 3-month history of a right breast mass. Fine needle aspiration of the mass was positive for malignancy. Physical examination, bilateral mammography and ultrasonography of both breasts revealed a 6-cm right breast mass associated with multiple enlarged axillary lymph nodes. However, metastatic work-up including chest X-ray, abdominal and pelvic ultrasonography, and bone scintigraphy demonstrated no abnormal findings. In addition, a complete blood count, renal function tests, serum electrolytes, liver function tests including AP, serum LDH, CEA and CA 15-3 were within normal limits.

According to the physical examination and metastatic work-up, she was diagnosed as having locally advanced infiltrating ductal carcinoma of the right breast. Therefore, she was initially treated with three cycles of cyclophosphamide, doxorubicin and 5-fluorouracil (CAF) neoadjuvant chemotherapy, which achieved a partial response. She was subsequently treated with modified radical mastectomy, which revealed a 3-cm grade II/III infiltrating ductal carcinoma. The surgical margins were clear and 4 out of 18 lymph nodes showed evidence of metastatic disease. She

Figure 1. Small, ovoid and circular sclerotic bone lesions in all tarsal, metatarsal and phalangeal bones of the foot. (A) Distal end of the femur. Proximal ends of the tibia and fibula. (B)

subsequently received three additional cycles of adjuvant CAF chemotherapy and adjuvant external-beam radiotherapy that consisted of 50 Gy in 25 fractions to the chest wall and regional nodes.

Twenty-eight months after initial treatment, at a routine follow-up visit, the patient complained of bilateral knee and foot pain. Plain X-rays showed multiple well-defined sclerotic lesions throughout the knees and feet (Figures 1A and 1B). According to the radiographic appearance, the most likely differential diagnoses included skeletal metastases of breast cancer and osteopoikilosis. Whole-body bone scintigraphy (Figure 2) showed no increase in uptake by the osteosclerotic lesions. In addition, there was no increase in serum LDH, CEA, AP or CA 15-3. We therefore diagnosed the patient's skeletal lesions as osteopoikilosis.

The patient's symptoms were relieved by nonsteroidal anti-inflammatory drugs and she was symptom-free for 15 months, when she developed progressive back pain. At that time thoracic spinal magnetic resonance imaging demonstrated a compression fracture of the 7th and 8th thoracic vertebral bodies, which caused cord compression (Figure 3). Whole-body bone scintigraphy showed increased uptake in the skull, ribs, left shoulder, thoracic and lumbar vertebral bodies, right humerus and left tibia (Figure 4). In addition, serum levels of LDH, AP and CA 15-3 were increased, but no increase in serum CEA was

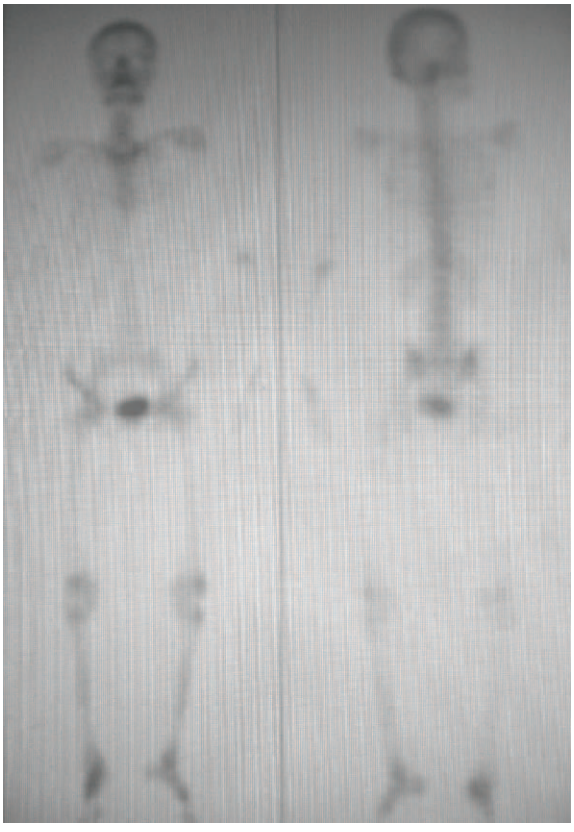

Figure 2. Whole-body bone scintigraphy showing no evidence of skeletal metastases.

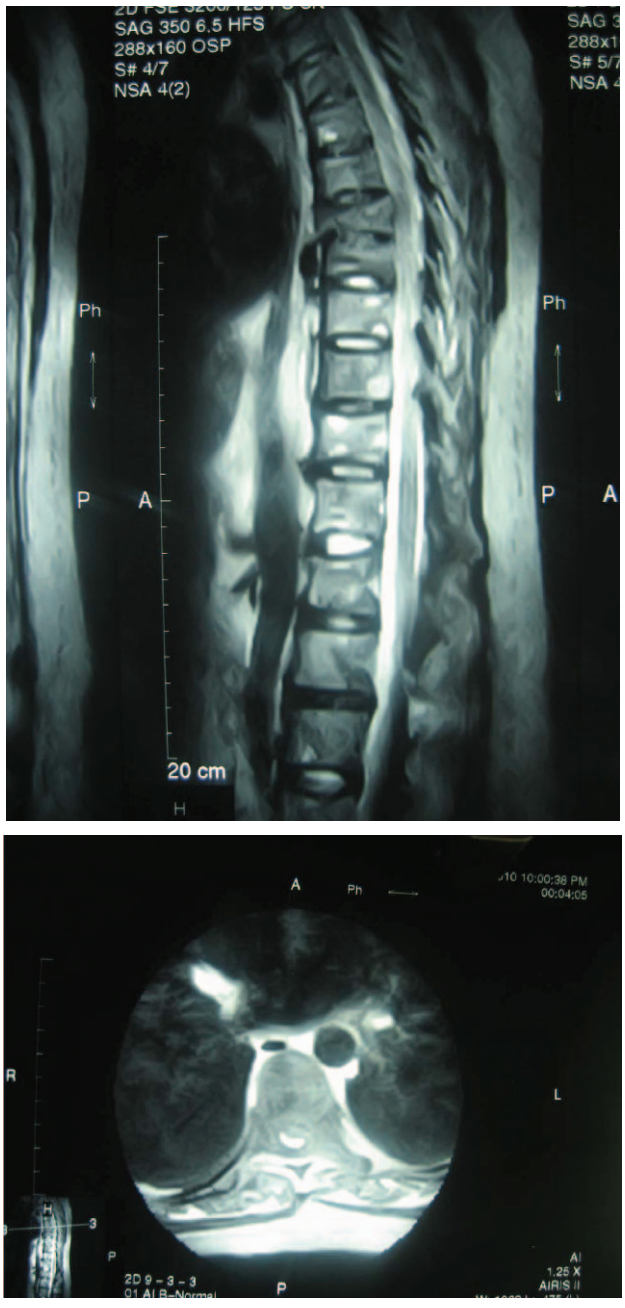

Figure 3. Sagittal (A) and cross-sectional (B) T2-weighted magnetic resonance imaging of the thoracic spine showing abnormal high-signal and compression fractures of the 7th and 8th thoracic vertebral bodies, causing cord compression. These lesions corresponded to metastatic disease.

noted. A plain X-ray of the pelvis revealed findings similar to those seen previously in her feet and knees (Figure 5). Therefore, the new spinal lesions were considered skeletal metastases and the patient was treated with surgery and palliative spinal radiotherapy for her rapidly progressive spinal cord compression. At the time of this writing she is well and currently receiving bisphosphonate therapy for her skeletal-related events.

Discussion

Skeletal metastasis is the most important differential diagnosis for bony lesions in patients with breast cancer, since the skeleton is the site of most distant failures in these patients. In addition, skeletal metastases are the main source of bone pain in these patients.[2] However, the possibility of rare primary underlying bone disorders such as osteopoikilosis or second malignancies such as plasmacytoma or multiple myeloma should be included in the list of differential diagnoses when bony pain is investigated, particularly when unusual sites such as the distal extremities are involved.[5,6] Less likely differential diagnoses for radiographic lesions in cancer patients include mastocytosis, tuberous sclerosis and synovial chondromatosis.[5]

Figure 4. Whole-body bone scintigraphy showing increased uptake in the skull, ribs, left shoulder, thoracic and lumbar vertebral bodies, right humerus and left tibia.

Figure 5. Plain X-ray of the pelvis showing numerous small, ovoid osteosclerotic lesions in both femoral heads, the ileum and around the acetabulum of the pelvis.

Concurrent osteopoikilosis and breast cancer may mimic skeletal metastasis in nonmetastatic breast cancer, as seen in our patient, or may mask skeletal metastatic foci in patients with skeletal metastatic breast cancer, as described by Ghandur-Mnaymneh et al.[7] In our literature review we found three previously reported cases of coincidental breast cancer and osteopoikilosis that mimicked[8] or masked[7,9] skeletal metastases.

The findings in our patient and literature review suggest that the radiographic appearance of osteopoikilosis should be taken into account, because this rare bone disorder may mimic or mask skeletal metastases in patients with breast cancer and thus lead to errors in the differential diagnosis.

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