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Osteoblastoma as a cause of osteomalacia assessed by bone scan

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A 27-year-old female patient was admitted to our hospital with a history of leg pain and mass. She had a benign osteoblastoma in right tibia. Resection of the tumor without treatment by vitamin D antagonist resulted in rapid cure of the osteomalacia. Bone scintigraphy with Tc-99m MDP revealed multiple hot uptakes in initial scan, and follow up scan showed a clear resolution of the lesions.

Key words: osteoblastoma, osteomalacia, bone scintigraphy

INTRODUCTION

OSTEOMALACIA secondary to tumor is rare and it can be cured after removal of the tumor. There have been several reports of osteomalacia associated with a tumor of bone or soft tissue.[1] We report a case of osteoblastoma patient who showed multiple focal areas of intense uptake on bone scintigraphy and this scintigraphic findings returned to normal on a repeated bone scintigraphy after resection of tumor.

CASE REPORT

A 26-year-old woman was admitted to our hospital with a two year history of difficulty in walking, weakness in right leg and pain with recent (2 months) swelling over the right calf. In the plain radiography, the cortex of the right tibia was destroyed with osteolytic lesion (Fig. 1 (A)). In MRI (enhanced-T1WI), outbuldging mass lesion with peripheral enhancement in diaphysis of right posterior tibial cortex (Fig. 1 (B)) is noted. In the initial bone scan, there were multiple hot uptakes in the right tibia, mandible, maxilla, both clavicles, left humerus, both ulna, posterior arc of right 7th to 9th and 12th ribs, L2, sacrum, right S-I joint, left proximal femur and calcaneus (Fig. 2). Plain radiography of these lesions in bone scan showed no abnormal findings (Fig. 3). We considered it as metastatic lesions or multiple myeloma, but the results of studies were negative. In the laboratory findings, the levels of serum calcium and inorganic phosphorous were low as 7.8 mg/d*l* (normal: 8.6–10.6) and 1.2 mg/d*l* (normal: 2.5–4.5). But the level of Alkaline phosphatase was high as 756 IU/*l* (normal: 80–270). Biopsy was done and it was revealed as osteoblastoma. After confirmed, the tumor was resected en bloc with wide margin. The multiple hot uptake lesions were considered as multiple osteomalacia lesions caused by osteoblastoma. In the 2 and 9 months follow up of the bone scan after resection of tumor, multiple hot uptake lesions were disappeared subsequently (Fig. 4) and the levels of serum calcium, inorganic phosphorous and alkaline phosphatase were recovered to normal (Table 1). The clinical symptoms were disappeared too without any further treatment with vitamin D.

DISCUSSION

The syndrome of tumor-induced osteomalacia was first reported by McCane in 1947.[1] It is a rare clinicopathological disorder with hypophosphatemia, hyperphosphaturia, low 1,25-$(OH)_2$ Vitamin D_3 and abnormalities in skeletal mineralization. Tumors combined with osteomalacia are Giant cell tumor, hemangiopericytoma, chondroblastoma, fibroma & fibroangioma, prostate cancer, osteosarcoma, MFH, chondrosarcoma, ETC and osteoblastoma etc. The pathogenesis of osteomalacia in osteoblastoma is not clearly defined. Humoral factors released by these tumors may impair proximal tubular functions such as

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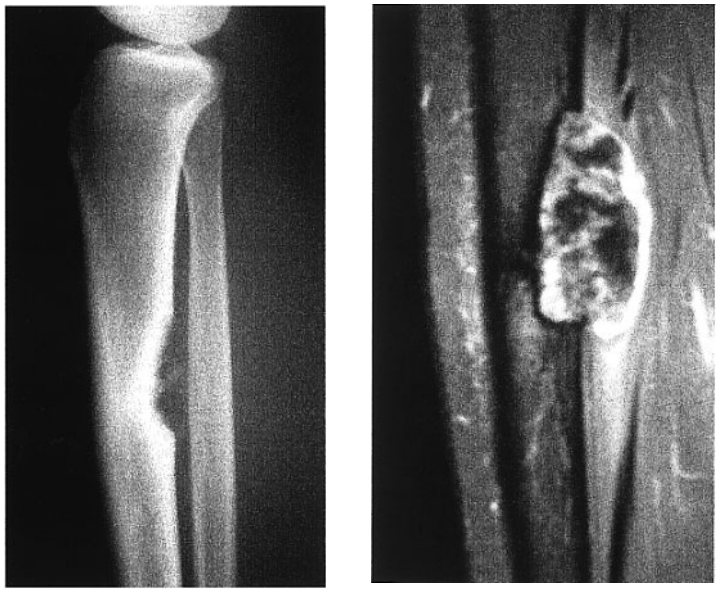


A B

Fig. 1 In the plain radiography, the cortex of the right tibia was destroyed with osteolytic lesion (A). In MRI (T1 weighted coronal image with enhancement), peripheral-enhanced outbuldging mass lesion is noted in diaphysis of right posterior tibial cortex (B).

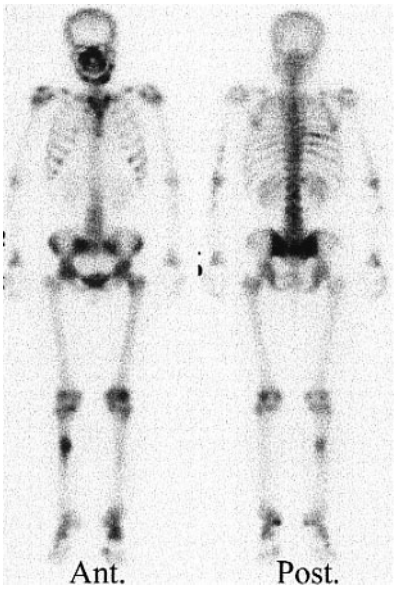


Fig. 2 In the initial bone scan, there were multiple hot uptakes in the right tibia, mandible, maxilla, both clavicles, left humerus, both ulna, posterior arc of right 7th to 9th and 12th ribs, L2, sacrum, right S-I joint, left proximal femur and calcaneus.

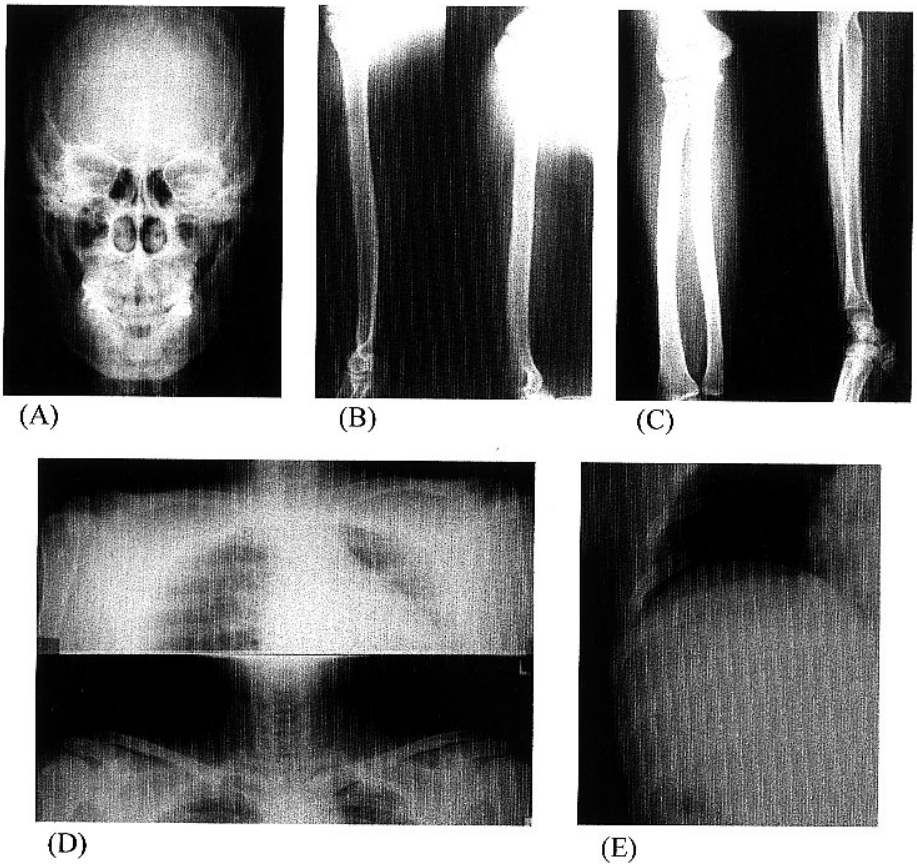
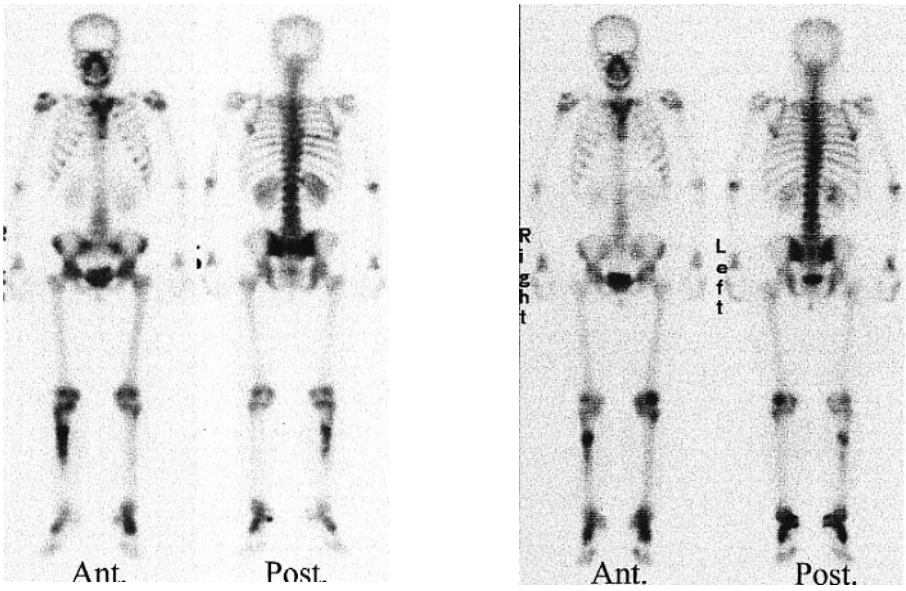


Fig. 3 Plain radiography of skull (A), left humerus (B), left forearm (C), both clavicle (D) and right rib series (E) showed no abnormal findings.



A B

Fig. 4 In the 2 (A) and 9 months (B) follow up of the bone scan after resection of tumor, multiple hot uptake lesions were disappeared subsequently.

Table 1 Changes of laboratory findings

	Initial	Post-op. 1 wk.	Post-op. 10 wks.	Post-op. 10 mons.
Ca (8.6–10.6 mg/d*l*)	7.8	8.3	8.9	9.6
P (2.5–4.5 mg/d*l*)	1.2	2.9	3.3	3.3
ALP (80–270 IU/*l*)	756	404	333	289

1α-hydroxylation of 25(OH)D and phosphate transport.[2] And there are several reports of hypothesis that cause osteomalacia in tumors are phosphatonin, PEX gene[3] and MEPE genes.[4]

Osteomalacia is a disorder in which mineralization of the organic matrix of the skeleton is defective. The presentation of osteomalacia in adults is usually more insidious and symptoms, when they occur, include diffuse skeletal pain and bony tenderness. The diagnosis of osteomalacia relies on abnormal biochemical findings and pseudofractures on radiographs and definite diagnosis rests on the biopsy. However, bone biopsy is an invasive technique and serum biological results in a diagnosis of osteomalacia is limited.[5] In osteomalacia, decrease in bone density is usually associated with loss of trabeculae and thinning of the cortices. The radiologic changes may be indistinguishable from those in osteoporosis. The specific finding that suggests osteomalacia is the presence of radiolucent bands. These radiolucent bands, called pseudofractures or Looser's zones, occur most often at sites where major arteries cross the bones and are thought to be due to the mechanical stress of the pulsation of these vessels. On radionuclide bone scans the pseudofractures appear as hot spots. Bone scan had been shown to play an useful role in the detection of osteomalacia.[6–8] Bone scan patterns are generalized as increased skeletal uptake, prominent costochondral junctions, increased periarticular uptake and focal hot uptake in the sites of pseudofractures. If pseudofractures are asymmetrically distributed, the scan findings mimics that of metastatic lesions. As in our ease, both symmetric and asymmetric and asymmetric focal hot uptakes were shown as reminiscent of bone metastases.

Bone scan is more sensitive in detecting pseudofractures than radiography.[6,7] As in our patient, many lesions are seen on bone scintigram, whereas normal in radiologically. Also, repeated bone scintigraphy after treatment (operation) should be done to differentiate between osteomalacia and bone metastases. In our case, repeated bone scan showed normalization of previous abnormal findings.

CONCLUSION

The combination of osteomalacia with a bone or soft tissue mass should raise suspicion of oncologic osteomalacia. Bone scan is useful for detection and follow-up after tumor resection in oncologic osteomalacia.

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