

## Periosteal osteoblastoma: Multimodal imaging of a rare neoplasm

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Osteoblastoma is a rare neoplasm, accounting for only 1% of the primary bone neoplasms. We report a case of periosteal osteoblastoma, a rare subtype of osteoblastoma. To the best of our knowledge, have only 30 cases of periosteal osteoblastoma appear in the English literature. Our case is that of a 41-year-old male with vague intermittent knee pain over a two-year period. Diagnostic imaging revealed an aggressive appearing lesion in the posterior distal femur, which was initially considered to be a surface osteosarcoma. Roentgenographic, CT, MRI, and bone scan features are presented.

### Introduction

Osteoblastoma is a rare benign tumor that accounts for fewer than 1% of all primary bone neoplasms. The neoplasm may be intramedullary, cortical, or subperiosteal in location (1, 2). Sixty to sixty-five percent of the osteoblastomas are cortical, and the remainder are medullary (3, 4). Subperiosteal location is very rare, with only 30 cases reported in the English literature (to the best of our knowledge and PubMed search). Osteoblastoma is a benign neoplasm that can be radiographically mistaken for several benign and malignant lesions, such as osteoid osteoma, aneurysmal bone cyst, infection, osteosarcoma, and metastasis (40). We present a case of periosteal osteoblastoma with radiographic evidence of bone formation, periosteal reaction, cortical disruption, and lucent region disrupting the medullary space, the cortex, and the periosteum. Radiographically, the lesion was considered to be a surface osteosarcoma.

### Case report

A 41-year-old man presented with intermittent left knee pain of two years' duration. The pain began after he fell performing a takedown maneuver during customs agent training. He began to have difficulty squatting fully. Over the two years, the pain had progressed to involve the hip and buttock. He developed occasional night pain, aching, stabbing and sharp sensations, and occasional knee instability. At presentation, he characterized the pain as 8/10 in severity, partially relieved with ibuprofen, and worsened by lying down. There was no pertinent past medical, surgical history, or family history.

Physical examination revealed a slender, well-appearing male. The left leg did not demonstrate overlying skin abnormalities, masses, or regional adenopathy. Light touch sensation was intact, and the site of knee pain was difficult to pinpoint. Vague discomfort was noted anteriorly and posteriorly. The patella tracked centrally and the knee was stable. Muscle strength was normal in flexion and extension.

Radiographs of the left knee showed a small mass at the posterior aspect of the distal femoral metaphysis. The mass was shaped like a dome and showed amorphous osteoid-type mineralization and aggressive periosteal reaction (Fig. 1). CT of the lesion showed that its epicenter was at the cortical surface, with a small mass projecting into the soft tissues and a lytic component that appeared to penetrating the cortex and extending into the medullary space (Fig. 2). Amorphous mineralization was present within the lesion, and aggressive periosteal reaction with sunburst morphology was present (Fig. 3). MRI showed the lesion straddling the cortex, with most of the tumor volume on the surface of

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Figure 1. 41-year-old male with periosteal osteoblastoma. An AP (top) and lateral (above) radiograph of the distal left femur demonstrate a lytic lesion with aggressive periosteal reaction.

the bone (Fig. 4). There was heterogeneous signal on T1- and T2-weighted sequences, with extensive surrounding marrow, periosteal, and soft-tissue edema (Fig. 5). The lesion was hot on radionuclide bone scan (Fig. 6). The radiologic impression was that of periosteal osteosarcoma.

The patient underwent an open biopsy. An intraoperative frozen section was obtained, which showed irregular small trabeculae of bone with osteoblastic cells lining the trabeculae and in the intertrabecular space mixed with occasional osteoclasts and some larger atypical cells. Due to the presence of some atypical cells, there was concern for osteosarcoma. The surgical site was closed in anticipation of preoperative chemotherapy. Further evaluation of the frozen and permanent histological sections demonstrated similar trabecular bone formation lined by plump osteoblasts and occasional osteoclasts with a richly vascular stroma typical of osteoblastoma (Fig. 7). There were occasional larger atypical epithelioid cells with abundant amphophilic cytoplasm and preserved nuclear-to-cytoplasmic ratios.

Such cells can be seen in osteoblastoma. The histopathological features in conjunction with the radiological findings supported the final pathologic diagnosis of periosteal osteoblastoma.

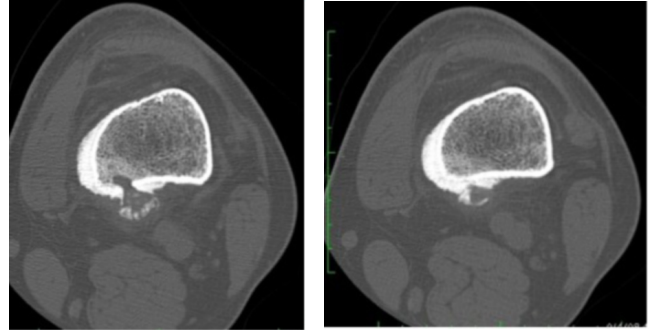


Figure 2. 41-year-old male with periosteal osteoblastoma. An axial CT shows cortical destruction with central osteoid mineralization. Solid periosteal reaction and focal area of aggressive periosteal reaction of sunburst pattern are also present.

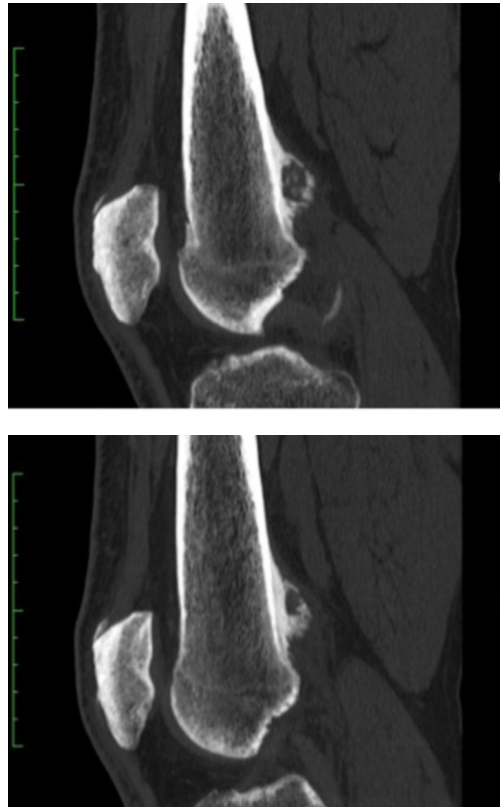


Figure 3. 41-year-old male with periosteal osteoblastoma. A sagittal reformatted CT also shows cortical destruction with central osteoid mineralization. Solid periosteal reaction and focal area of aggressive periosteal reaction of sunburst pattern are also present.



Figure 4. 41-year-old male with periosteal osteoblastoma. Sagittal T1-weighted MR image demonstrates T1 hyperintensity and cortical disruption.

The patient subsequently underwent radical resection of the lesion with cryotherapy. Reconstruction of the surgical defect was accomplished with internal fixation using a cortical strut allograft placed over morsellized cadaveric allograft. The pathology of the resection specimen was again consistent with osteoblastoma, subperiosteal type. Importantly, there was no evidence of invasion of the underlying bone, further helping to exclude osteosarcoma (Fig. 8). The surgical margins were free of neoplasm.

### Discussion

Osteoblastoma occurs most commonly in the second and third decades of life, with cases reported ranging from six months to 75 years of age (1, 4). More than 90% of osteoblastomas occur before the age of 30 (5), with a mean age of 20.4 years (6). Males are affected twice as frequently as females (ratio of 2:1). Clinically, pain is the most common presenting feature, and diagnosis may be delayed up to two years from the onset of symptoms (7, 5, 4, 1). Aggressive forms of osteoblastoma may present with more severe pain, which may be due to localized tissue destruction (8). The pain from osteoblastoma may be more severe at night and may not respond to aspirin or nonsteroidal anti-inflammatory drugs (7, 8). McLeod et al reported pain relief in only 7% of patients in their series of 123 patients (3). Osteoblastomas have a predilection for the posterior elements of the spine and the diaphysis and metaphysis of long bones of the appendicular skeleton (1, 4). Jackson et al reported 35% percent of osteoblastomas in the spine, 10% in the femur, and 5% in the tibia in a series of 184 patients (5). Lucas et al reported 32% in the vertebral column, 12% in the femur, and 10% in the tibia (4).



Figure 5. 41-year-old male with periosteal osteoblastoma. Axial (top), coronal (middle), and sagittal (above) long TR with FS images show perilesional edema. The lesion is mixed high and low in signal intensity.

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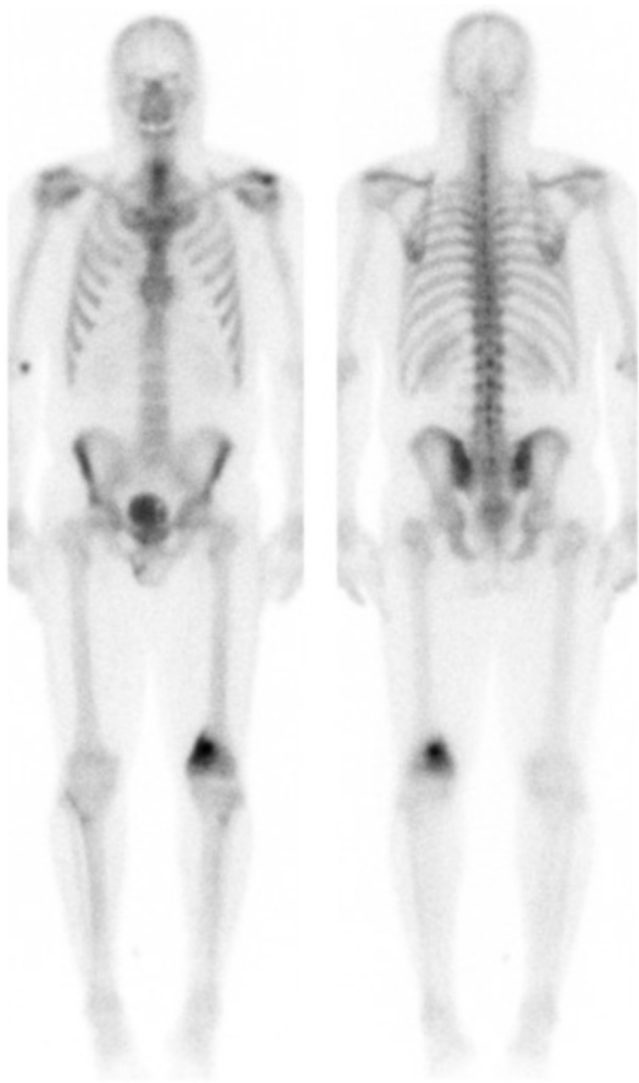


Figure 6. 41-year-old male with periosteal osteoblastoma. Anterior and posterior whole-body bone scans show increased uptake in the distal femur lesion.

Of the long-bone lesions, 42% were metaphyseal, 36% were diaphyseal, and 22% were within the epiphysis (4). Analyzing the previously reported periosteal osteoblastomas, Mortazavi et al reported that more than half of the cases were of the long bones, which is not the case with conventional osteoblastoma. There have been nine femur, seven cranial/facial, four humerus, three rib, two tibia, two fibula, one radius, and one scapula cases of periosteal osteoblastomas (2) in addition to the femur lesion being reported here.

On imaging, osteoblastoma may be difficult to distinguish from osteoid osteoma. Generally, lesions less than 1.5 cm are classified as osteoid osteoma; those greater than 1.5 cm are classified as osteoblastoma (9, 4). As in our case, osteoblastoma can be difficult to distinguish from osteosarcoma.

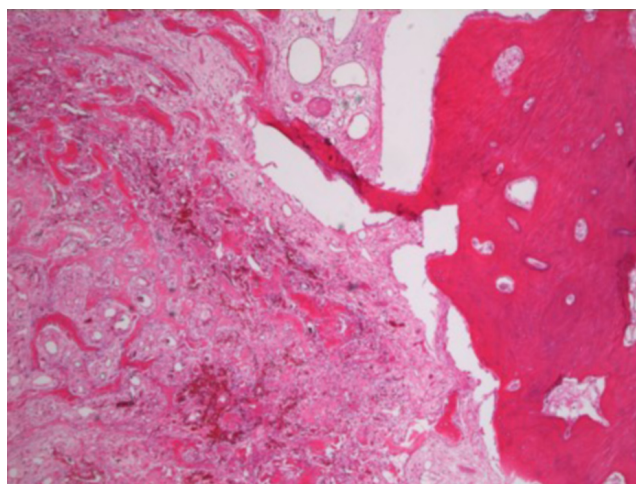


Figure 7. 41-year-old male with periosteal osteoblastoma. Bone forming lesion of the surface of the cortex (left) with no invasion of the underlying cortex (right).

The imaging findings in our case are mostly consistent with those of other reported periosteal osteoblastomas (6, 10, 2, 11, 12). Although our case also demonstrates appearance of cortical destruction with extension of mass into the medullary space, on histopathology, the mass was noted to not invade the underlying cortex (Fig. 7). Sulzbacher et al also reported cortical erosion, although small (measuring only 5

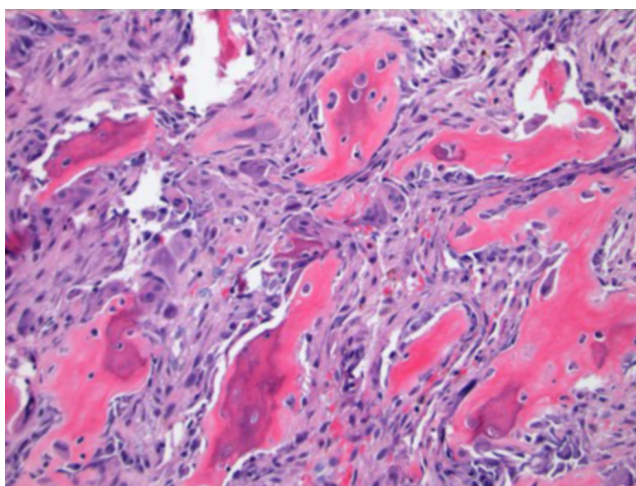


Figure 8. 41-year-old male with periosteal osteoblastoma. Irregular and variably mineralized small trabeculae of woven bone lined by osteoblasts and occasional osteoclasts. A fibrovascular stroma and osteoblasts are seen in the intertrabecular space. A few of the osteoblasts are enlarged with abundant cytoplasm (epithelioid osteoblasts).

mm [12]). The case reported by Kawaguchi et al demonstrated low signal intensity on long TR (6); however, our

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case and the case reported by Nakatani et al show high and low signal on long TR sequences (11).

At histology, most osteoblastomas show irregular trabeculae of woven bone surrounded by a single row of osteoblasts set in a loose connective tissue stroma. There may be variable amounts of bone mineralization, and there is a highly fibrovascular stroma in the intertrabecular space (5, 4). Occasional osteoclasts are present as well. Mitotic figures may be present, suggestive of high growth potential (5). Large epithelioid osteoblasts (as seen in this lesion) can be seen in 24% of cases (4).

Osteoblastomas may be treated by wide excision or curettage; however, incomplete resection may sometimes result in recurrence (3). Recurrence rates have been reported between 10% and 20% (7, 9, 4).

In conclusion, osteoblastoma is a benign primary bone neoplasm that presents with pain. Osteoblastomas have a variable radiologic appearance ranging from indolent to very aggressive. Cure is achieved with wide resection, and a 10% to 20% recurrence rate has been associated with curettage. Periosteal osteoblastoma is a rare subtype of osteoblastoma with only 30 (including our case) instances in the English literature.

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