

Periosteal Osteosarcoma of the Fifth Metatarsal: A Rare Pedal Tumor

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ARTICLE INFO

Level of Clinical Evidence: 4

Keywords:

foot
metatarsal
neoplasm
sarcoma
surgery

ABSTRACT

Periosteal osteosarcoma is a rare pedal chondroblastic osteosarcoma that rarely involves the medullary bone. In the present report, we describe the case of a woman who presented with a periosteal osteosarcoma localized to her left foot.

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Periosteal osteosarcoma (POS) is a rare chondroblastic osteosarcoma with known radiologic and histopathologic features (1). This tumor is a cartilage-rich, juxtacortical tumor with a good prognosis (1,2). It tends to arise from the diaphyseal surface of the tibia and femur (1,3), is most prevalent in young adults, and is classified as a low- to intermediate-grade surface osteosarcoma (4). The purpose of the present report is to describe an exceedingly rare case of POS localized to the foot in a woman. To the best of our knowledge, this is the first reported case of POS in the foot. Furthermore, medullary involvement is also very rare in association with POS (1), and we believe the present case represents the 23rd case of POS with medullary involvement to be described in the peer-reviewed data.

Case Report

A 33-year-old Azari woman presented to our hospital on referral for evaluation and treatment of a painful mass localized to the dorsolateral surface of her left foot of approximately 6-months' duration. She had no history of foot trauma, and her medical and family histories were unremarkable. The physical examination revealed a firm, fixed, tender mass on the dorsolateral aspect of her

left fifth metatarsal. No evidence was found of ipsilateral inguinal or popliteal adenopathy. Plain radiographs of her left foot showed a coarse, perpendicular periosteal reaction with Codman's triangle and thickening of the fifth metatarsal cortex, without diaphyseal scalloping or erosion (Fig. 1). The radiographs also suggested the presence of a sunburst appearance of cortical expansion, and the lesion decreased in intensity from the cortical base (medullary surface) toward the surface. The cortex of the fifth metatarsal bone was intact, and cortical scalloping was not seen. Magnetic resonance imaging in the coronal view showed a mass with a low signal intensity on the T₁-weighted image and high signal intensity on the short T₁-weighted inversion recovery sequence images localized to the fifth metatarsal and compatible with the areas of periosteal reaction identified radiographically, reflecting the high water content of the bone lesion. Secondary bone marrow involvement was also evident as a high signal intensity in the medullary cavity (Fig. 2). The laboratory data, including complete blood count, erythrocyte sedimentation rate, calcium, phosphorus, and alkaline phosphate levels were normal.

Because of our concern for osteosarcoma, the extent of the neoplasm (coursing from the proximal to distal metaphysis), the expansive nature of the cortical involvement, and the rapid growth of the lesion and associated pain, we recommended radical excision of the entire fifth metatarsal. This option was reviewed with the patient, who agreed to this treatment course, which would yield an accurate diagnosis and could likely be curative. The patient subsequently underwent surgery under general anesthesia and in the supine position. Using tourniquet hemostasis (without compression

Financial Disclosure: None reported.

Conflict of Interest: None reported.

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Fig. 1. Plain radiograph showing sunburst periosteal reaction in lateral surface of fifth metatarsal bone.

exsanguination), the full length of the fifth metatarsal, from the proximal to distal articular surfaces, effecting an approximately 5-mm safe margin, was excised. The adjacent tendons and interphalangeal muscles were normal in appearance and were preserved. The entire specimen was submitted in formalin for pathologic examination. The peroneus brevis tendon was anchored to the distal margin of the cuboid using an intraosseous nonabsorbable suture, and the wound was closed in anatomic layers. The foot was dressed in a sterile bandage and stabilized with a below the knee posterior splint.

After surgery, the patient experienced an unremarkable post-operative recovery. Histopathologic examination revealed bony

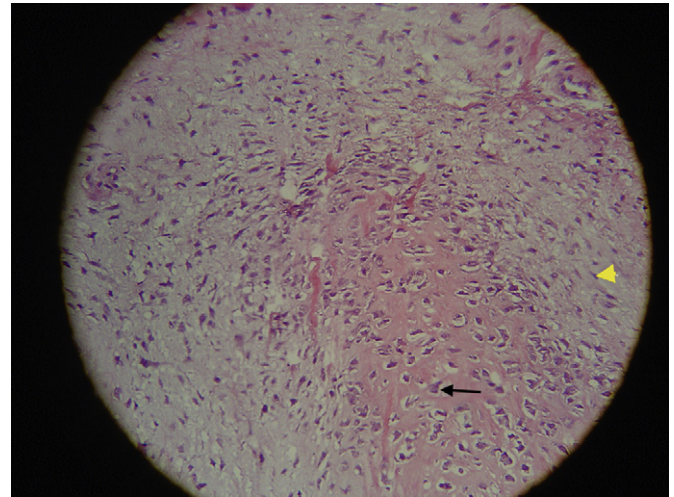


Fig. 3. Neoplastic proliferation of spindle cells (arrowhead), cartilaginous component (arrow), and osteoid formation (magnification 4×, hematoxylin and eosin stain).

trabeculae with neoplastic proliferation of spindle cells, a cartilaginous component with a gradual blending pattern, and a few areas of calcified chondroid matrix and atypical cartilage (Fig. 3). Some atypical large hyperchromatic pleomorphic cells with osteoid formation were also seen (Fig. 4). The tumor grade was 1 (localized, without spreading), and the bone marrow was not involved. The pathologic specimen was sent for consultation to 2 other experienced pathologists, both of who concurred with the diagnosis. By 3 weeks post-operatively, the patient had resumed use of regular shoes, and by 4 weeks, she had resumed regular weight bearing activities without the use of a brace or any form of walking aid. Because of the pathologic diagnosis of POS, the patient underwent an oncologic evaluation, chest radiography, and abdominal ultrasonography. The findings from the latter imaging modalities were normal. Thus, it was determined that she did not require adjunct chemotherapy or radiotherapy, although close clinical surveillance was in order. A postoperative follow-up radiograph was obtained 6 months after surgery and did not show any evidence of tumor recurrence (Fig. 5), consistent with her clinical progress. At the last follow-up visit, 9 months after excision of the fifth metatarsal neoplasm, the patient displayed no

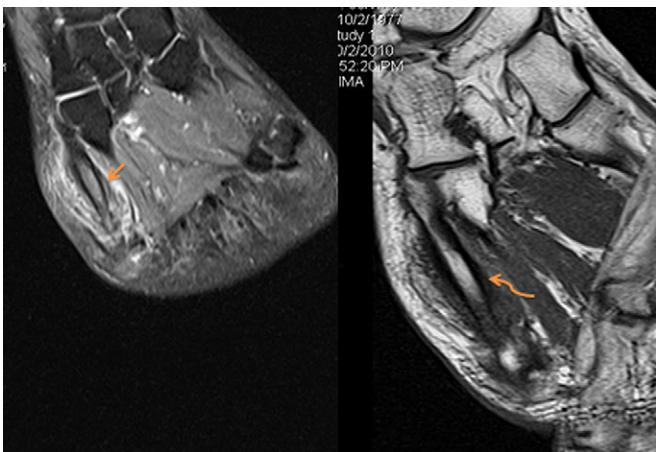


Fig. 2. Axial magnetic resonance image showing soft tissue with intermediate signal intensity around fifth metatarsal bone and high signal intensity within bone marrow.

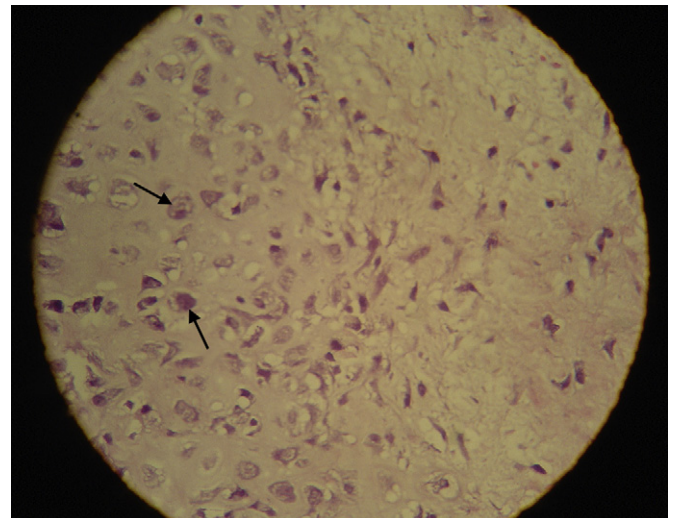


Fig. 4. Some atypical large hyperchromatic pleomorphic cartilage cells (arrows) (magnification 10×, hematoxylin and eosin stain).



Fig. 5. Plain radiograph showing postoperative changes (absent fifth metatarsal) without any recurrence.

evidence of recurrence and was ambulating without difficulty in regular shoe gear. She was subsequently scheduled for ongoing follow-up at 6-month intervals, and ongoing follow-up clinical and radiographic re-evaluation on a regular basis for the initial 5 post-operative years was planned.

Discussion

Three types of surface, or juxtacortical, osteosarcoma have been reported: parosteal, periosteal, and high-grade surface osteosarcoma (HGSO) (4). POS is a rare chondroblastic bone-forming tumor that was first described by Unni et al. It was differentiated from parosteal osteosarcoma in 1976 (5). Previous studies have demonstrated its predominance in patients younger than 20 years old (3); the female/male ratio is 3:2 (6). Patients typically present after a few weeks or months of pain, swelling, and tenderness, with or without a visible or palpable mass. The most common location of POS, as for conventional osteosarcoma, is the knee (1,5). On imaging, POS appears as an elongated, partially mineralized mass on the cortical surface of a long bone in the diaphyseal region, with a thickened underlying cortex and a solid periosteal reaction at the margins (4). The characteristic radiographic findings of POS consist of a broad-based soft-tissue mass attached to the cortex (100%), cortical thickening (82%), extrinsic scalloping of the cortex (92%), and a periosteal reaction (95%) (7). Periosteal osteosarcomas are much less dense radiographically than parosteal osteosarcomas, reflecting the difference in their histologic differentiation. They also occur in an older group of patients compared with POS and typically have a narrow base, with or without a distinct cleavage plane. Radiologically, periosteal chondrosarcoma, Ewing's sarcoma, parosteal osteosarcoma, and HGSO constitute the

differential diagnosis for POS (1). HGSO is generally considered to be the closest radiographic and clinical "look-alike" to POS, in that both lesions are typically diaphyseal, peripherally based, and commonly manifest a substantial periosteal reaction. The appearance on plain film radiographs might be indistinguishable from that of a typical medullary osteosarcoma. However, computed tomography and magnetic resonance imaging often demonstrate that the HGSO lesion typically encircles the entire bone, but POS does not. POS commonly presents with cortical thickening and scalloping and reactive marrow edema, not seen with HGSO. POS also commonly shows low attenuation on computed tomography and a high signal intensity on T₂-weighted magnetic resonance imaging, reflecting the high water content of these largely chondroblastic lesions. In general, HGSO does not show these same findings on computed tomography and magnetic resonance imaging, although POS appears as a broad-based soft tissue mass attached to the cortex, just as does HGSO.

POS generally conveys a better prognosis than does other forms of osteosarcoma (2). In the present case, once the diagnosis of POS was determined, an oncology consultation and co-management were undertaken, and no adjunct chemotherapy or radiotherapy was administered.

We treated our patient with wide, local excision, because medullary involvement was present, even though such involvement is considered rare in POS (1,5). Our review showed only 23 previously reported cases of POS with medullary involvement; thus, we believe the tumor we have described in our patient is rare. Moreover, only 6 other cases of POS with secondary bone marrow involvement have been described in previous reports (1,8,9). Finally, we are not aware of any other published reports of POS localized to the foot. From our understanding of the published data and our experience with the present patient, we believe that POS localized to the foot is a rare disease that can be treated with wide excision and regular follow-up examinations.

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