# Parosteal Osteosarcoma of the 2nd Metatarsal

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# Abstract

While masses of the foot are relatively common, bone forming lesions of the foot are less common. The differential diagnosis includes benign and malignant lesions. A thorough history and physical examination, along with selective imaging, can guide the practitioner to an accurate diagnosis. For malignant lesions, appropriate local and whole body imaging consistent with published national guidelines in order to stage the patient's disease helps to guide treatment. Knowledge of the national history of the lesion will guide decision making for appropriate surgical resection and the need for any adjuvant therapy. Postresection surveillance of malignant or locally aggressive lesions is also important in the patient's postoperative care. We present a case of parosteal osteosarcoma of the 2nd metatarsal.

**P**arosteal sarcoma is a rare primary malignant tumor of bone. The neoplastic cells of parosteal osteosarcoma form osteoid from the surface of the bone. This tumor has also been termed juxtacortical osteogenic sarcoma, parosteal osteogenic sarcoma, ossifying parosteal sarcoma, and par or periosteal ossifying fibrosarcoma. The tumor is most commonly located around the knee followed by the proximal humerus. In previous large clinical series, the incidence of parosteal sarcoma is between 1.5% and 4% of all primary bone tumors.<sup>1,2</sup> It is most common in the third decade of life.<sup>3-5</sup> Typically low grade malignancies, parosteal sarcomas have a more favorable prognosis than periosteal (intermediate grade) or high grade surface osteosarcoma.<sup>1</sup>

There have been 6 previous reports of parosteal sarcoma in the foot. In a 1997 report from the Sloan-Kettering database,<sup>4</sup> 1 of 24 cases was reported in the foot and the patient went on to local recurrence and an amputation below knee at 6 years. In a report of 25 cases from the Enneking experience,<sup>6</sup> the authors describe a single case involving the 5th metatarsal treated with complete ray resection and had no evidence of disease at 11 years. In a report of 64 cases of parosteal osteosarcoma from the Latin American Registry of bone pathology,<sup>1</sup> 2 cases of the foot were noted without further detail about the treatment or patient outcomes. A case report from the radiology literature was of the 2nd metatarsal and was treated with a complete ray resection and had no clinical recurrence of the grade I tumor at 18 months.<sup>7</sup> The final case in the literature involved a 19-year-old male with a lesion in the 4th metatarsal, which was treated by en bloc resection of the 4th and 5th metatarsals with no evidence of disease at 22 months.<sup>8</sup> In these 6 cases, 4 were treated with complete ray resection and 2 were treated with amputation.

We present a case of parosteal osteosarcoma of the 2nd metatarsal. The patient provided written informed consent for print and electronic publication of this case report.

# **Case Report**

Our patient is a 51-year-old woman with a 1-year history of right foot pain and a palpable mass on her foot. She was previously seen by a podiatrist for her foot pain and difficultly with shoe wear. Plain radiographs were obtained (Figure 1A-C), a mass was noted, and she was referred to an orthopedic oncologist. She had no history of trauma to the foot and reported 10/10 pain of her right foot. Upon review of symptoms, she had no history of fevers, chills, night sweats, nausea, or emesis.

Figure 1. Anteroposterior radiograph (A), oblique radiograph (B), and lateral non-weight bearing radiograph (C), obtained from podiatrist's office.



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Figure 2. Two sequential axial cuts from a bony CT scan. This demonstrated a surface lesion arising from the 2nd metatarsal head/neck region.



Figure 3. A 3-dimentional CT reconstruction of the mass.

She had anemia of unknown etiology, dyspnea on exertion, migraine headaches, and low back pain attributed to a prior work injury.

Past medical history included hypercholesterolemia, arthritis, and migraine headaches. The patient was not taking any medications. Social history was positive for 1 pack-per-day tobacco use.

Physical examination demonstrated a slender woman who appeared her stated age in no acute distress. Her examination was negative for lymphadenopathy, abdominal masses, and any range of motion limitations. Focused examination of the right lower extremity demonstrated 2+ dorsalis and posterior tibial pulses, sensation intact to light touch of the plantar/dorsum/1st web space of the foot. The patient had 5+/5 strength in all muscle groups. She had a palpable mass in the 1st-2nd intermetatarsal space, which was firm and non-mobile. The mass



Figure 4. A coronal T1-weighted MRI on the left demonstrating the mass without connection to the medullary canal and a T2 section without any medullary edema on the right.

was palpable primarily dorsally. There was divergence of the hallux from the lateral 4 rays and hallux valgus on examination. The patient had negative clinical staging for metastasis.

The patient had preoperative radiographic staging, which included a chest x-ray, whole body bone scan, and computed tomography (CT) scan of the chest. A chest x-ray was obtained in the office, which demonstrated 2 small lesions consistent with calcified granulomas. The CT scan demonstrated several mildly enlarged lymph nodes with the largest being 1.8 cm x 1.1 cm (Figures 2, 3). The whole body bone scan was negative for other sites of disease or metastasis. The preoperative staging imaging was negative for metastatic disease. The patient was clinically and radiographically staged as Stage IA, T1N0M0 in accordance with the American Joint Commission on Cancer (AJCC) 7th edition Staging Manual.9 An operative planning magnetic resonance imaging (MRI) scan was obtained. The MRI demonstrated 4 cm x 3 cm x 3 cm mass arising from the 2nd metatarsal head and neck region (Figure 4). There was no connection with the medullary canal found on MRI. Thickening of the lateral cortex of the 1st metatarsal suggested pressure changes and a non-acute nature to the mass.

# **Differential Diagnosis**

#### Extraskeletal Osteochondroma

This is a benign soft tissue lesion that can be commonly found in the foot. It usually presents as a calcified mass on plain radiograph and is most common in the skeletally mature. The peak incidences are in the third and sixth decades of life.<sup>10,11</sup> Advanced imaging studies usually demonstrate a soft tissue mass adjacent to the bone with calcifications without associated bony destruction. On MRI they are typically well demarcated, associated with a tendon and near a joint, but not associated with a joint. This tumor has not shown malignant transformation, but does have a local recurrence rate of up to 18%.<sup>12</sup> This lesion is less likely in our patient because on imaging it does arise from the bone with associated changes of the bony architecture.

#### **Bizarre Parosteal Osteochondromatous Proliferation**

Bizarre parosteal osteochondromatous proliferation (BPOP), also known as a Nora lesion, due to its first description by Nora and colleagues<sup>13</sup> in 1983, is also a rare lesion with less



Figure 5. A non-weight bearing AP radiograph of the foot at the 1-month postoperative visit. Note the intermetatarsal screw to serve as the transverse intermetatarsal ligament.



Figure 6. Gross resection specimen showing a cortically based lesion.

than 200 cases described in the literature. This lesion is a combination of bone cartilage and fibrous tissues. It

usually has an exophytic shape and most commonly arises from the proximal and middle phalanges, and the metatarsal and metacarpal bones. The hands are 4 times more commonly affected than the feet. The lesion can also be seen in skull, long bones, maxilla, and even in the sesamoid bones of the foot. It most commonly affects patients in the third and fourth decades of life with males and females being equally affected. This is a benign condition, which is locally aggressive. The radiological findings are similar to our case presentation in that CT scans usually show intensely calcified masses with well, defined margins. However, with BPOP there is usually no disruption of the underlying bones and the adjacent soft tissues are normal. On MRI, the lesions are isointense with muscle on T1 and have high intensity rims with low intensity core on T2 weighted images. Our patient was within the appropriate age distribution, location of lesion, and similar radiographic appearance to BPOP.

#### **Tumoral Calcinosis**

Characterized by calcification in periarticular areas, tumor calcinosis, is more commonly associated with major joints such as the hip, shoulder, knee, and elbow, but has been described in the foot.<sup>14,15</sup> The mass is usually painless and occurs before the age of 20 in most cases. It can be further classified into primary or secondary, if the patient has predisposing conditions such as chronic renal disease, hyperparathyroidism, malignancy, sarcoidosis, hypervitaminosis D, milk-alkali syndrome, massive osteolysis, scleroderma, or pseudoxanthoma elasticum. Plain radiographs demonstrate oval-shaped or round, well-defined lesions with the pathognomonic "chicken-wire" calcifications. CT scans in the more mature lesions have well defined cortical margins and no trabecular structures. MRI demonstrates low intensity of T1 and T2 imaging due to the calcium present. Our patient did not have abnormal levels of calcium and her lesion was less well defined on plain radiograph and advanced imaging. Our patient was also older than the typical demographic for this lesion.

#### Heterotopic Ossification

Our patient had no history of trauma to the foot.

**Stress Fracture that Healed With Exuberant Callus** Our patient had no history of repeated weight-bearing activity other than walking and there was not evidence of a stress reaction on plain radiographs or CT scan.

# Treatment

After discussion at an institutional, multidisciplinary Bone and Soft Tissue Tumor conference, a recommendation was made for partial versus complete ray

resection. The patient was treated with partial second ray resection. A lag screw was placed from the 1st to the 3rd metatarsal in order to prevent progression of hallux valgus, (Figure 5), after the intermetatarsal ligament was resected with the mass. The gross specimen followed by microscopic views can be seen in Figures 6-8.



Figure 7. Anastomosing trabeculae of bone with hypocellular spindle cell stroma (Hematoxylin and eosin [H&E] x200).



Figure 8. High power highlighting spindled cells, which exhibit minimal atypia (H&E x400).



Figure 9. A clinical photograph of the patient's foot at the 6-month postoperative visit with a well-healed incision.

The patient is still being followed in the orthopedic oncology clinic with serial chest x-rays and clinical examinations of the foot. Further imaging to the foot will only be performed if there is a change in clinical symptoms, such as pain, or the presence of a palpable mass.

Currently, the patient is 2 years post-resection, walking without assistive devices or modified shoe wear, and has no evidence of disease. A clinical photograph of the patient's foot was obtained (**Figure 9**). The patient continues to have hallux valgus, but given her ability to fit into shoes and her lack of pain, we will continue to manage this conservatively.

## Discussion

Masses of the foot can have a broad differential diagnosis including both benign and malignant neoplasms of bone. Given the possibility of malignancy, a systematic approach must be taken. A thorough history and physical examination must be performed initially with attention to age and medical comorbidities. Additional specific questioning of the presence of night pain, length of time the mass has been present, change in size of the mass, and pain profile is helpful. Plain radiographic imaging of the foot must be obtained followed by axial imaging of the lesion. If an aggressive bone neoplasm remains in the differential chest, imaging should be obtained in accordance with National Comprehensive Cancer Network (NCCN) guidelines.<sup>16</sup>

A systematic approach to the patient with a foot mass including a clinical history and physical examination, selective use of imaging, and discussion at a multidisciplinary institutional tumor board can lead to the correct diagnosis and optimal patient management.

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