Mixed Cavernous and Capillary Intraosseous Hemangioma Of the Foot

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Hemangiomas of bone are rare lesions accounting for approximately 1% of all primary bone tumors. Intraosseous hemangiomas of the foot are especially rare, with only sparse reports in the literature. Presented here is a case study of an erosive bony lesion of the midfoot that was microscopically and histopathologically proven to be a mixed cavernous and capillary hemangioma. Eradication of the lesion during diagnostic biopsy obviated further treatment.

Osseous hemangiomas are rare benign tumors. They account for approximately 1% of all primary osseous neoplasms.1-3 Hemangiomas of osseous structures are most commonly reported in the vertebral column and the calvaria.3-5 They are found uncommonly in flat and tubular bones.2, 3, 6 Rarely have osseous hemangiomas been found in the foot.

Dorfman et al7 reported intraosseous hemangiomas of the foot in two locations, the calcaneus and the navicular bone, in 1971. In 1989, Murari et al8 reported three cases of intraosseous hemangiomas: two metatarsal and one calcaneal. Mirra9 reported two cases occurring in tarsal bones of the foot. Two cases, both talar, were reported by Sheth et al4 and Wu10 in 1994.

Osseous hemangioma presents with localized dull, aching pain that may progress over several months.4, 10, 11 Soft-tissue swelling and a growing mass have been reported.5, 11, 12 History of trauma does not appear to be a predisposing factor.5, 11,13 Hoffmann and Israel5 reported a range of incidence of 30 to 60 years of age; this is consistent with additional reports of peak incidence of 40 years of age7, 13 and 50 years of age.4, 9 Women are more frequently affected than men; various female-to-male ratios have been reported, including 2:17, 14 and 4:1.5, 13

Intraosseous hemangioma presents radiographically as lytic solitary lesions with or without a sclerotic rim.4, 8, 10, 13 The lytic area may have a honeycomb trabecular pattern caused by an expanding tumor or a sunburst pattern, with striations radiating from the center.5, 7, 13 The tumor is usually well demarcated. Endosteal cortical scalloping, cortical erosion with absence of hyperostosis, and a reactive shell of periosteal bone have also been reported.11-13 Bone scans show evidence of slight to moderate uptake of radionuclide.12 There may be gradually increasing uptake within the slowly flowing blood of the hemangioma.13 Magnetic resonance imaging shows a homogeneous low-intensity signal on T1-weighted images and a high-intensity signal on T2-weighted images.11, 13 These findings are nonspecific. Computed tomographic findings confirm the pathology of plain radiographs.11 Angiography is not frequently reported. Peterson et al12 and Wold et al5 noted that there is a type of intraosseous hypervascular lesion in which feeding arteries and draining veins may be dilated but are of normal shape and contour.

Microscopically, hemangiomas can be divided into three types: capillary, cavernous, and mixed cavernous...
Capillary hemangiomas are composed of multiple thin-walled capillaries that usually are filled with blood and separated by scant connective-tissue stroma. The lumina may be organized and partially or completely thrombosed. The capillaries are arranged in lobules or radiating loops. Capillary hemangiomas are composed of large, cavernous vascular spaces that may be filled with blood and are separated by scant connective-tissue stroma. The blood-filled sinusoid channels form a growing mass that erodes or displaces the normal tissue. Stress created by these lesions contributes to the development of bony trabeculae in the lesion secondary to osteoblastic reinforcement and remodeling by osteoclast. Both capillary and cavernous hemangiomas are unencapsulated and well defined.

Differential diagnoses in cases of osseous lesions of the foot should include aneurysmal bone cyst, cortical chondroma, subperiosteal giant cell tumor, cortical osteofibrous dysplasia, eosinophilic granuloma, cholesteatoma, multiple myeloma, metastatic carcinoma, osteoma, Paget’s disease, and hyperparathyroidism.

Case Study

A 57-year-old man was initially seen with a chief complaint of pain and swelling in the medial dorsal aspect of his right foot. The patient complained of an aching pain of 3 weeks’ duration that increased during activity. There was no history of related trauma. On examination, there was localized tenderness of the dorsal first and second metatarsocuneiform joints and the naviculocuneiform joint. This was exacerbated with motion. The patient had a medical history consistent with gout, rheumatoid arthritis, intermetatarsal neuroma, herniation of disks L4-5 and L2-3, bulging of disks L3-4 and L5-S1 with resultant sciatica and degenerative joint disease, and diverticular colon with colostomy repair. The patient was currently taking probenecid and indomethacin. Social review was negative for tobacco use since 1980 and positive for occasional alcohol consumption. The patient was mildly overweight.

Radiographic examination revealed bony destruction of the naviculocuneiform joint and the first, second, and third metatarsocuneiform joints (Fig. 1). Magnetic resonance imaging with sagittal T1- and T2- and axial T1-weighted images of the spine revealed asymmetric polyarthritis, suggesting ankylosing spondylitis. Magnetic resonance imaging with axial T1-weighted short tau inversion recovery, sagittal and coronal proton density, and T2-weighted images was performed on the right foot. These studies showed marked erosion and deformity of the cuneiform bones, the naviculocuneiform joints, and the bases of the second and third and, to a lesser extent, the fourth metatarsals, as well as mild erosion of the cuboid (Fig. 2). There was granulation tissue around all of these joints, but it was most pronounced around the cuneiforms. Additionally, there was mild-to-moderate generalized subcutaneous tissue edema (Fig. 3). Focal edema was noted in the proximal shaft of the third metatarsal. There was also prominent edema within the sinus tarsi. A whole-body technetium-99m methylene diphosphonate bone scan revealed increased uptake in the cervical thoracic junction, costovertebral area, and mid and lower lumbar spine. There was also intense increased uptake of the right midfoot (Fig. 4 A-C).

A complete blood count, sedimentation rate determination, and biochemical and rheumatoid profile were ordered. Laboratory serologic tests for diabetes, gout, rheumatoid arthritis, and ankylosing spondylitis were negative. The only abnormalities noted were elevated blood urea nitrogen (29 mg/dl), cholesterol (230 mg/dl), GGT (112 U/liter), and alanine aminotransferase (87 U/liter). The patient was then scheduled for biopsy.

Surgical Presentation

A dorsal incision centered just lateral to the dorsalis pedis and anterior tibial artery was made. Dissection

Figure 1. Dorsoplantar radiograph showing erosion of the metatarsocuneiform joints, the naviculocuneiform joint, and the cuboid.
was carried through the subcutaneous tissues to the level of the intercuneiform and intermetatarsal space between the first and second and between the second and third cuneiforms. Biopsy of the dorsal aspect of the metatarsal and cuneiform was performed. The metatarsal and cuneiform base area was noted to be replaced by fibrous tissue. The bone did not have a normal trabecular or cortical appearance. Anaerobic and aerobic cultures revealed no growth of organisms. Microscopic examination showed a bony erosion and a proliferation of small capillary or slightly dilated cavernous vascular spaces. These were lined by a single layer of endothelium, without significant pleomorphism, solid proliferating areas, or anaplasia. The bony trabeculae were occupied by the hemangioma and loose connective tissue separating the bland vascular space. A few osteoblasts and osteoclasts were noted lining the trabeculae (Fig. 5 A and B). Ragged edges were noted in the eroded bone. The tumor appeared to affect the interface between bone and the adjacent cartilage. A diagnosis of a benign tumor consistent with an osseous hemangioma, mixed cavernous and capillary type, was made.

Postoperatively, the patient was maintained non-weightbearing to the right foot with crutch-assisted ambulation. The patient experienced limited postoperative pain and only minimal edema. The postoperative course was without complication. The patient was referred to an expert on embolization of hemangiomas for angiographic examination and destruction of the intraosseous hemangioma. Preprocedure magnetic resonance imaging, Doppler ultrasonography, and arteriography showed no sign of the tumor.

Specifically, magnetic resonance T1- and T2-weighted proton density, gradient echo, and fat-suppressed fast spin echo T2-weighted images showed no evidence of arteriovenous or venous malformation. There was no evidence of bony infarction, muscle masses, or tumor. Doppler venous noninvasive vascular examination revealed neither deep venous thrombosis nor vascular malformation. Arteriography revealed occlusion of the dorsalis pedis with major flow through the posterior tibial artery, no evidence of vascular malformation, and no arteriovenous fistula or malformation.

Further surgical intervention was therefore deemed unwarranted, and the patient was maintained for 6 weeks in a walking cast. Follow-up radiographs revealed a prominent first cuneonavicular area with profound arthritic changes but showing improvement since biopsy.

**Discussion**

Historically, vascular anomalies were treated initially with surgical proximal arterial ligation. However, with reconstituted arterial inflow of the nidus by neovascular recruitment and evidence of recurrent macrofistulous feeders from microfistulous connections, this treatment option often yielded suboptimal results. Embolotherapy has emerged as the primary therapeutic modality in the management of vascular anomalies owing to the difficulty of eradicating the nidus of these lesions and the improvement of embolic agents and catheter delivery systems. Often vascular anomalies are located in inaccessible or difficult-to-
reach areas. Therefore, reliance on the endosurgical skill of the interventional radiologist has increased. One mode of treatment is a percutaneous puncture technique, which allows direct access to the vascular lesion. This direct attack with an endovascular ablative agent does not affect the arterial inflow system or the capillary bed, minimizing tissue loss.16

In this presented case, preprocedure magnetic resonance imaging and ultrasonography showed that embolization therapy was not warranted. Magnetic resonance imaging and ultrasonography showed no evidence of arteriovenous or venous malformation, and structures were normal, without bony infarction. It was believed that during biopsy the blood supply to the tumor had been cut off, leading to eradication of the tumor, and that the bone was now healing.

Figure 4. Technetium-99m methylene diphosphonate scan of the lower extremity. A, Immediate flow indicating increased uptake in the midfoot region. B, Blood-pooled phase showing continued uptake in the midfoot region. C, Two-hour delayed phase indicating intense uptake consistent with bone destruction.

Figure 5. Photomicrographs of an intraosseous hemangioma, magnification ×10 (A) and ×20 (B). Note the proliferation of small capillary and dilated cavernous vascular spaces.
Eighteen months after biopsy, there has been no recurrence of hemangioma, and the patient's foot is asymptomatic.

The patient was subsequently diagnosed with carcinoma of the liver that was unrelated to the diagnosis of intraosseous hemangioma. The tumor was determined to be nonresectable, and the patient died of the disease 2 years later.

**Conclusion**

Intraosseous hemangioma is a rare, benign bone tumor accounting for less than 1% of all osseous neoplasms.\(^1\)\(^-\)\(^3\) Although it is most commonly seen in the vertebral column and the cranium, it should always be considered in the differential diagnosis of tumor presentations in the foot.\(^5\)\(^-\)\(^6\) Lesions are usually asymptomatic; when they are painful, however, surgical excision or endosurgical vascular ablative therapy may be warranted.\(^6\)\(^-\)\(^16\)

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**References**


**Additional References**

