Orthopaedic Case of the Month

Ankle Pain and Swelling in a 23-year-old Man

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History and Physical Examination

A 23-year-old man referred by his primary care physician presented with a 1.5-year history of progressive left ankle pain and swelling. He recalled no history of trauma other than stepping on a horseshoe crab when he was 8 years old and having the tail removed in a local emergency department. He denied recent fevers, chills,

night sweats, weight loss, or weight gain. Physical examination revealed substantial nonpitting edema around the hindfoot region extending up the distal 1/3 of his leg. There was no skin discoloration. He had mild tenderness to palpation along the peroneal and posterior tibial tendons. There was normal strength and sensation in the foot and leg. No lymphadenopathy was noted. His laboratory values, including complete blood count, erythrocyte sedimentation rate (ESR), C-reactive protein (CRP), uric acid, rheumatoid factor, and Lyme titer were all within normal limits. Imaging studies, including MRI (Fig. 1), CT scan (Fig. 2), and bone scan (Fig. 3), were performed. Based on the history, physical examination, laboratory tests, and imaging studies, what is the differential diagnosis?

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Each author certifies that all investigations were conducted in conformity with ethical principles of research and per discussion with the internal review board, formal processing was unnecessary. This work was performed at Beth Israel Deaconess Medical Center.

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Imaging Interpretation

Sagittal T1-weighted (Fig. 1A), sagittal T2-weighted fat-suppressed (Fig. 1B), and axial T1-weighted fat-suppressed contrast enhanced (Fig. 1C) MR images of the ankle showed a T1 hypointense and T2 hyperintense soft tissue mass remodeling the undersurface of the sustentaculum tali. There was a small irregular central area of low signal consistent with calcification.

Coronal (Fig. 2A) and sagittal (Fig. 2B) reformatted CT images of the ankle showed an area of smooth osseous remodeling at the inferior aspect of the sustentaculum tali. There was a 7- × 4-mm triangular calcification adjacent to the area of remodeling.

Radionuclide bone scan (Fig. 3) of the bilateral feet and ankles showed radiotracer uptake in the left posterior calcaneus.

Fig. 1A–C (A) Sagittal T1-weighted and (B) T2-weighted fat-suppressed MR images of the ankle show a T1 hypointense and T2 hyperintense soft tissue mass remodeling the undersurface of the sustentaculum tali. There is a small irregular central area of low signal

consistent with calcification. (C) An axial T1-weighted fat-suppressed contrast-enhanced MR image of the ankle shows enhancement of the mass and surrounding edema.

Fig. 2A–B (A) Coronal and (B) sagittal reformatted CT images of the ankle show an area of smooth osseous remodeling at the inferior aspect of the sustentaculum tali (arrows). There is a 7- × 4-mm triangular calcification adjacent to the area of remodeling.

Fig. 3 A radionuclide bone scan of the bilateral feet and ankles shows radiotracer uptake in the left posterior calcaneus.

Synovial sarcoma
Vascular malformation

A CT-guided biopsy of the patient's left pericalcaneal mass was performed for histologic and microbiologic analyses (Fig. 4).

Based on the history, physical examination, imaging studies, and histology, what is the diagnosis and how should this lesion be treated?

Histology Interpretation

The histologic specimen from the core biopsy contained fibroadipose tissue and skeletal muscle with reactive myofibroblastic proliferation and admixed chronic inflammation (Fig. 4); there was no histologic evidence of malignancy.

Differential Diagnosis

Foreign-body pseudotumor
Myositis ossificans

Diagnosis

Foreign-body pseudotumor

Fig. 4 A photomicrograph shows fibroadipose tissue with reactive myofibroblastic proliferation and dense chronic inflammation (Stain: hematoxylin and eosin; original magnification: $\times 400$).

Discussion and Treatment

This patient presented with an unusual soft tissue mass causing bony remodeling of the underlying calcaneus. In general, soft tissue masses of the foot are fairly common. Eight percent of benign and 5% of malignant soft tissue tumors are located in the foot and ankle [13].

Penetrating injuries have long been reported to cause pseudotumor reactions when near bone [4, 9, 11, 16]. These injuries are most commonly reported in the hand, with the foot as the second most common site. Thorns and wooden splinters are the most frequent etiology [6]. Of the 31 cases of pseudotumor resulting from thorn, wood, or plant splinters, 19 had osteolysis only, five had periosteal reaction without osteolysis, and seven had a combination of the two [9]. Smooth remodeling of the adjacent bone, as seen in this case, is suggestive of a more chronic and indolent process.

Foreign-body reactions vary in time to symptoms from days to years and a prior event may not be remembered [17]. Foreign bodies can be retained for a significant time before diagnosis [1]. Additionally, the duration of symptoms of patients with tumor or tumor-like lesions of the foot can vary dramatically. A study of 83 patients found symptoms varied from 1 month to 12 years for benign lesions and 1 month to 9 years for malignant lesions [12]. In our patient, the foreign body had been retained for a previously unreported duration of time, 15 years, with symptoms of 1.5 years. The invaluable piece of information in identifying this mass before definitive treatment was the patient's history of stepping on a horseshoe crab tail as

a child. The event happened so many years before his symptoms that initially he did not recall it. Only with careful questioning did the inciting event come to mind. This emphasizes the importance of a thorough history [20].

The use of MRI, CT, and ultrasound has increased the detection of soft tissue masses and foreign bodies [3, 7, 17]. Initial radiographs can have limited utility in this workup unless the foreign body is radiopaque. On MRI, the composition of the mass and reactive changes in the bone and adjacent soft tissues were nicely observed. Finally, the foreign body, given its mineralization, was well delineated with CT. Identifying a surrounding rim of fluid-rich granulation tissue or a fluid-filled cyst is an important feature of foreign-body reaction/pseudotumor [10, 15]. CT-guided biopsy has an accuracy of 90% for musculoskeletal lesions when compared with results found on open biopsy [21] and was helpful in this case to ensure this was not a malignant lesion before definitive treatment.

Myositis ossificans is soft tissue calcification often resulting from trauma. It progresses over a period of weeks to months and is characterized by a central cellular area surrounded by an intermediate zone of osteoid formation with a peripheral shell of well-organized bone [2]. The MR appearance of myositis ossificans varies as the lesion matures. Early lesions are poorly defined, isointense to muscle on T1-weighted images, and heterogeneously hyperintense on T2-weighted images and have diffuse surrounding soft tissue edema. As peripheral calcification develops, peripheral low signal may become visible on MRI. Mature lesions are well-defined masses isointense to fat centrally and low signal peripherally, on T1- and T2-weighted images, without surrounding soft tissue edema [14]. In contrast, the lesion in our patient was characterized by central calcification with surrounding soft tissue reaction and no well-organized bone.

Synovial sarcoma occurs most frequently in young adults and is the most common sarcoma of the foot [19]. It is a malignant mesenchymal tumor with variable but demarcated epithelial and mesenchymal spindle cell differentiation [8]. These cells stain positive for cytokeratin, epithelial membrane antigen, and bc12 protein but negative for CD34 [8]. Greater than 90% are associated with the chromosomal translocation $t(X:18)(p11.2;q11.2)$ [8, 18]. MRI classically shows a well-defined nodular mass with intermediate signal intensity on T1 and high signal on T2 sequences with heterogeneous contrast enhancement [22]. Approximately $\frac{1}{3}$ of synovial sarcomas have calcification in the sarcomatous tissue [8], which somewhat resembled our patient's imaging. However, there were no spindle cells in the patient's biopsy specimen and his MRI showed central calcification with surrounding, somewhat diffuse reactive tissue, not the distinct irregular calcifications more typically seen in synovial sarcoma.

Vascular malformations often develop in embryonic life and may enlarge with time. They consist of variably sized vascular channels lined by cytologically bland endothelial cells that show expression of CD34 and CD31 by immunohistochemistry. They may be visible on radiography if calcifications are present and are characterized on MRI by low signal intensity on T1 and high signal on T2 with flow voids [5]. The calcification in our patient's lesion was triangular and not typical of the phleboliths seen in vascular malformations. Also, the reactive tissue around the calcification was more diffuse and ill-defined than is typically the case in a vascular malformation. The histologic analysis confirmed this was not a vascular malformation.

The patient was taken to the operating room to remove the foreign body and débride the reactive tissues. Dissection was taken down to a distended pseudocapsule overlying the foreign body. On opening this pseudocapsule, purulent fluid came forth and was sent for culture. The foreign body was located and grossly on inspection was consistent with the tail of a horseshoe crab. The sclerotic cavity was curetted thoroughly and the wound copiously irrigated. The surgical pathology specimen consisted of two fragments that measured 0.7 cm each. One of these fragments contained a slender piece of foreign material that showed a single pointed end. Surrounding this foreign material was a zone of necrosis with admixed fibrinopurulent exudate. Directly apposed to this necrotic area was an area of dense fibrosis, reactive myofibroblasts, mild chronic inflammation, and abundant admixed hemosiderin-laden macrophages (Fig. 5). Again, no histologic evidence

Fig. 5 A photomicrograph shows foreign material consistent with a horseshoe crab tail. Directly surrounding this foreign substance is a zone of necrotic tissue with admixed neutrophilic and histiocytic inflammation. Adjacent to this necrotic material is a region of dense fibrosis, reactive myofibroblastic proliferation, chronic inflammation and hemosiderin-laden macrophages (Stain: hematoxylin and eosin; original magnification: $\times 400$).

of malignancy was identified. Culture specimens obtained during surgery grew no bacteria and stains for fungi and mycobacterium were negative.

At 1 month followup, the patient's wound broke down. Culture specimens were obtained and grew *Pseudomonas*, a possible dormant infection. He returned to the operating room for irrigation and débridement. Repeat culture specimens grew coagulase-negative *Staphylococcus* and diphtheroids. He was treated for a polymicrobial infection with vancomycin and Zosyn[®] (Wyeth, Madison, NJ) for 6 weeks with resultant healing of the wound. He had one additional bout of swelling and pain in this same area of his foot 2 months later requiring another operation, but cultures from the second procedure were negative. As a precaution, he was treated for an additional 6 weeks with IV antibiotics and the wound responded clinically very rapidly. Since that time, he has had no additional episodes of swelling, pain, or wound breakdown for more than a year, at the time of this writing, and he has returned to all activities.

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