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Case Report

Multifocal subacute osteomyelitis in adjacent bones in the ankle without septic joint

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ABSTRACT

Osteomyelitis is an inflammation involving bone and/or bone marrow most often the result of bacterial infection of the bone. In children, osteomyelitis most often has an acute presentation and is caused by hematogenous spread. When osteomyelitis is seen in the extremities, conventional radiography is the first-line imaging modality preformed for diagnosis with magnetic resonance imaging employed for further delineation or as a problem-solving tool. A healthy 6-year-old female presented with a history of nonspecific left leg pain for 3-5 weeks which gradually progressed to focal left ankle pain and swelling. Further workup revealed multifocal subacute osteomyelitis with Brodie's abscesses seen on imaging in the absence of a septic joint. This was an uncommon presentation for the following reasons: Patients with multifocal osteomyelitis usually present in the acute setting, as opposed to the subacute setting. When osteomyelitis is multifocal or Brodie's abscesses are present adjacent to the joint capsule, concomitant septic joint is commonly seen.

Introduction

Osteomyelitis is defined as inflammation involving bone and/or bone marrow. Although many infectious and noninfectious etiologies may result in osteomyelitis, it is almost exclusively the result of bacterial infection of the bone [1,2]. Bacteria may reach the bone through a number of pathways: direct inoculation from traumatic wounds, adjacent tissue affected by cellulitis or septic arthritis, or through hematogenous seeding. Osteomyelitis can be classified as acute, subacute, or chronic infection based on the time between the onset of symptoms and diagnosis [1,3]. Acute osteomyelitis is diagnosed within 2 weeks of the onset of symptoms. Subacute osteomyelitis is diagnosed 2 weeks after the onset of symptoms. Chronic osteomyelitis is diagnosed months after the onset of symptoms [1,3].

In children, osteomyelitis most often has an acute presentation and is caused by hematogenous spread [4]. However, these distinctions are not absolute; osteomyelitis in children may have any classification and reach the bone through any pathway [1,4]. When osteomyelitis is seen in the extremities conventional radiography is often the initial image modality

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used during diagnosis with subsequent magnetic resonance imaging (MRI) employed for further evaluation and/or surgical planning [5–7]. Notably, when using radiographs, the typical findings of osteolysis, periosteal reaction, and sequestra may not appear until 2 weeks after the onset of infection [8].

Case report

A healthy 6-year-old female who participates in recreational gymnastics presented with a history of nonspecific left leg pain for 3–5 weeks which gradually progressed to focal left ankle pain and swelling. This was in the absence of any known inciting injury. Several weeks prior she had a bout of diarrhea and fever with a headache for 24 hours but had remained afebrile since. Due to her symptoms, she began to modify her activity and eventually stopped gymnastics due to pain. On physical exam, she had point tenderness just superior to the lateral malleolus. Her exam was otherwise unremarkable. Laboratory evaluation including C-reactive protein, erythrocyte sedimentation rate, and complete blood count with differential were unremarkable.

Ankle radiographs showed multiple lucencies along the distal aspect of the left tibial metaphysis and distal fibula (Fig. 1) with diffuse soft tissue swelling around the left ankle and a small left ankle joint effusion. With the given clinical

Fig. 1 – Frontal radiograph of the left ankle shows multifocal, periphyseal, osteolytic lesions (arrows) **involving the distal tibial and fibular metaphysis.**

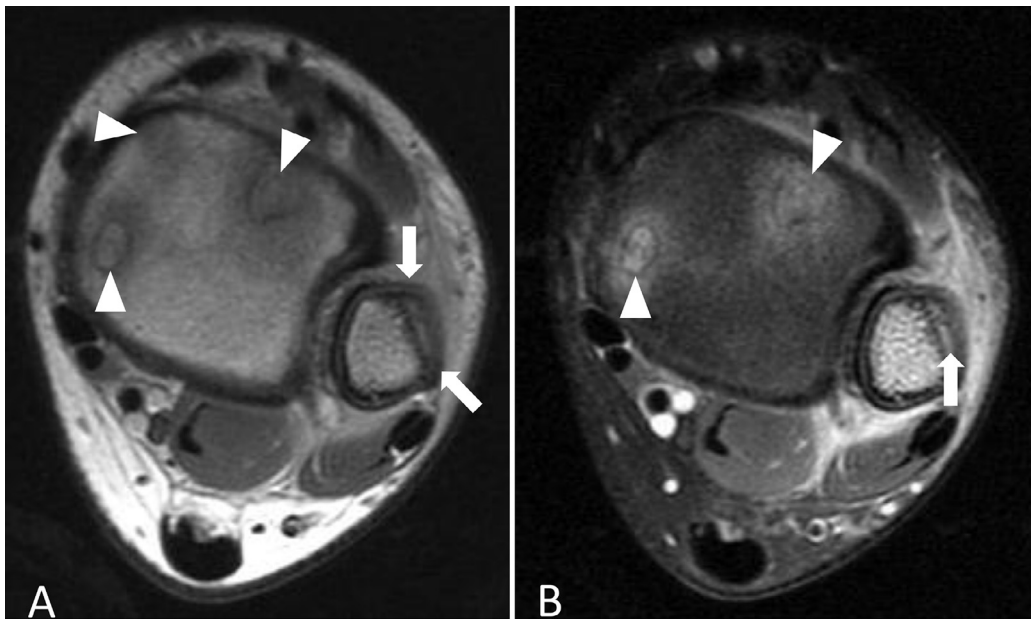

Fig. 2 – (A) **Axial T1-weighted image without fat-saturation demonstrates focal lesions within the distal tibial metaphysis** (arrowheads) **with hypointense rims and central hyperintensity consistent with internal proteinaceous fluid. Adjacent confluent, marrow replacing T1 hypointensity. Additionally, there is permeation of the adjacent fibular metaphyseal cortex from the 12 o'clock to 4 o'clock positions** (arrows) **with a lenticular-shaped, hyperintense collection in the subcortical marrow space. Findings are consistent with osteomyelitis and multifocal Brodie's abscesses.** (B) **Axial, fat-saturated, T2-weighted image demonstrates the focal Brodie's abscesses in the distal tibial** (arrowheads) **and fibular metaphysis** (arrow)**. Patchy adjacent marrow edema, notably, more pronounced in the distal fibula suggesting the tibial infection is less acutely inflamed relative to the fibula. Patchy soft tissue edema adjacent to the tibia and fibula, again, more pronounced and likely more acute about the fibula.**

Fig. 3 – Sagittal T1-weighted image without fat-saturation demonstrates confluent marrow replacing T1 hypointensity (arrows) in the distal tibial metaphysis adjacent to the periphyseal abscess (arrowhead). There is an absence of joint effusion with preservation of the fat contours anterior and posterior to the tibiotalar articulation.

Fig. 4 – Sagittal, fat-saturated, T1-weighted image postgadolinium intravenous contrast material administration shows patchy enhancing tibial metaphyseal marrow edema and inflammatory edema in the soft tissues, anteriorly. The abscess within the distal tibial metaphysis (arrow) has intrinsic T1 hyperintensity consistent with proteinaceous material and was shown not to have associated enhancement when compared to precontrast material administration T1-weighted images (not shown).

history, infection/septic joint was suspected, but further imaging with MRI was recommended.

An MRI of the ankle was subsequently performed which demonstrated multiple abscesses in the distal tibial and fibular metadiaphyses along the physes (Figs. 2-4). Adjacent to each abscess was confluent, marrow-replacing, T1 hypointensity (Figs. 2A and 3), consistent with MRI findings of osteomyelitis in this patient with multiple Brodie's abscesses. Notably, there was a lack of ankle effusion (Fig. 4), effectively ruling-out septic arthritis. In patients aged 0-18 months, it is common for the infection to cross the physis due to transphyseal vessels. However, in children aged 2-16 the physis becomes relatively avascular and less likely to become infected, as seen in this patient [9].

The following day the patient underwent fluoroscopic guided aspiration of the distal fibular metaphyseal fluid collection by the orthopedic team for cultures prior to starting antibiotics. Aspirate revealed a small amount of bloody fluid. The other 2 fluid collections in the distal tibia were not aspirated. She was then started on oral Cephalexin for 6 weeks. Bacterial cultures, fungal smear, fungal cultures, acid fast smear, mycobacterial cultures, and Lyme serology were ultimately all negative.

She was followed with serial laboratory evaluation which normalized. After completion of antibiotics she had no residual pain or tenderness and was able to resume normal activity with no restrictions. Repeat radiographs were scheduled but not obtained due to appointment cancelation.

Discussion

This case demonstrates a unique presentation of subacute osteomyelitis in a child with multifocal disease and Brodie's abscesses in 2 adjacent bones in the absence of a septic joint.

Subacute osteomyelitis occurs in approximately 5 in 100,000 children per year in high-income countries, with studies suggesting it may have a higher prevalence in low- and middle-income countries [10]. While acute osteomyelitis typically present with anemia, leukocytosis, elevated erythrocyte sedimentation rate, and elevated C-reactive protein, [11,12] subacute osteomyelitis can be difficult to diagnose because the disease has an insidious onset, presents with milder symptoms, and typically lacks the classical systemic reaction [13,14].

Multifocal osteomyelitis occurs rarely, with studies showing only 10% of children who present with osteomyelitis have multiple foci of infection [12,15]. Patients with multifocal osteomyelitis usually present in the acute setting with a hematogenous spread [12,16], as opposed to the subacute setting seen in this case. We initiated a literature review in PubMed using the keywords: subacute, osteomyelitis, and multifocal, excluding Chronic Recurrent Multifocal Osteomyelitis, in subjects between 0 and 18 years of age. This

search produced one relevant case reports from 1976 [17] suggesting the uniqueness of this kind of presentation.

When osteomyelitis is multifocal or involves segments of the bone adjacent to the joint capsule, concomitant septic joint is commonly seen [18,19]. Additionally, intraosseous infected fluid collections called Brodie's abscesses can be seen as a subacute manifestation of osteomyelitis and septic joint [20]. In contrast, noninfectious chronic recurrent multifocal osteomyelitis typically does not present with fluid collections or Brodie's abscess [21]. The typical radiographic appearance of chronic recurrent multifocal osteomyelitis is a lytic lesion that develops progressive sclerosis and hyperostosis over time [22]. The patient in this case presented with multifocal infection around the ankle joint with Brodie's abscesses seen on imaging. However, this was in the absence of a septic joint making the presentation uncommon.

Despite orthopedic aspiration, culture-negative osteomyelitis, as seen in our case, is a common occurrence in both unifocal and multifocal disease. The actual rates of culture-negative osteomyelitis occurrence vary with studies reporting anywhere between 15% and 61% [12,23-25]. This usually delays diagnosis until after lytic bone lesions, like Brodie's abscess, have occurred and can be seen on imaging [13].

We presented an unusual case of subacute multifocal osteomyelitis in adjacent bones of the ankle in the absence of septic joint. These findings are unique and warrant consideration by radiologists and treating clinicians when evaluating atypical joint pain.

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