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Acute Multifocal Hematogenous Osteomyelitis in a 13 year-old: A Case Report

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Background: *Acute hematogenous osteomyelitis (AHOM) is a serious pyogenic infection that is generally caused by bacteria and is most commonly found in children. A multifocal presentation of the disease is rare, mostly seen in the newborn and only in a small percentage of older children. We report a case of multifocal AHOM in a previously healthy, 13-year old girl, who presented herself with several painful and swollen joints, accompanied by high fever. The history, clinical course, radiologic findings and management rationale are presented in this report.*
Methods: *A review of the clinical, laboratory, radiological and microbiological data of the patient was done.*
Results: *Arthrocentesis of a swollen ankle, done under suspicion of septic arthritis, yielded clear and sterile synovial fluid. Magnetic resonance imaging scans of affected joints later confirmed AHOM close to the joint, causing the sterile joint effusion.*
Discussion: *A lesson to be learned from this case is that in case of a clinically suspected septic arthritis, a negative joint puncture could mean an osteomyelitis close to the joint.*

Keywords: AHOM, pediatric, child, acute, multifocal, hematogenous, osteomyelitis, staphylococcal

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Acute hematogenous osteomyelitis (AHOM) is a serious pyogenic infection that is generally caused by bacteria and is most commonly found in children.[1] Usually, antibiotics and the careful use of surgery can effectively treat the disease, but when managed poorly it can be life threatening, or at best, a debilitating and crippling illness.[2] AHOM is frequently caused by S. aureus, which is responsible for over 80-90% of culture positive cases.[5] These cases amount to 20-90% of all cases.[1]

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AHOM typically presents with local pain and fever that has lasted for approximately 3 days on presentation in the hospital.[3] It is most commonly found in tubular bones. The femur and tibia are affected in one third of the cases, followed by the humerus.[4,5] Infection of pelvic bones, which was seen in the following case, is rare.[6] Multifocal presentation of acute hematogenous osteomyelitis occurs even less frequently in older children.

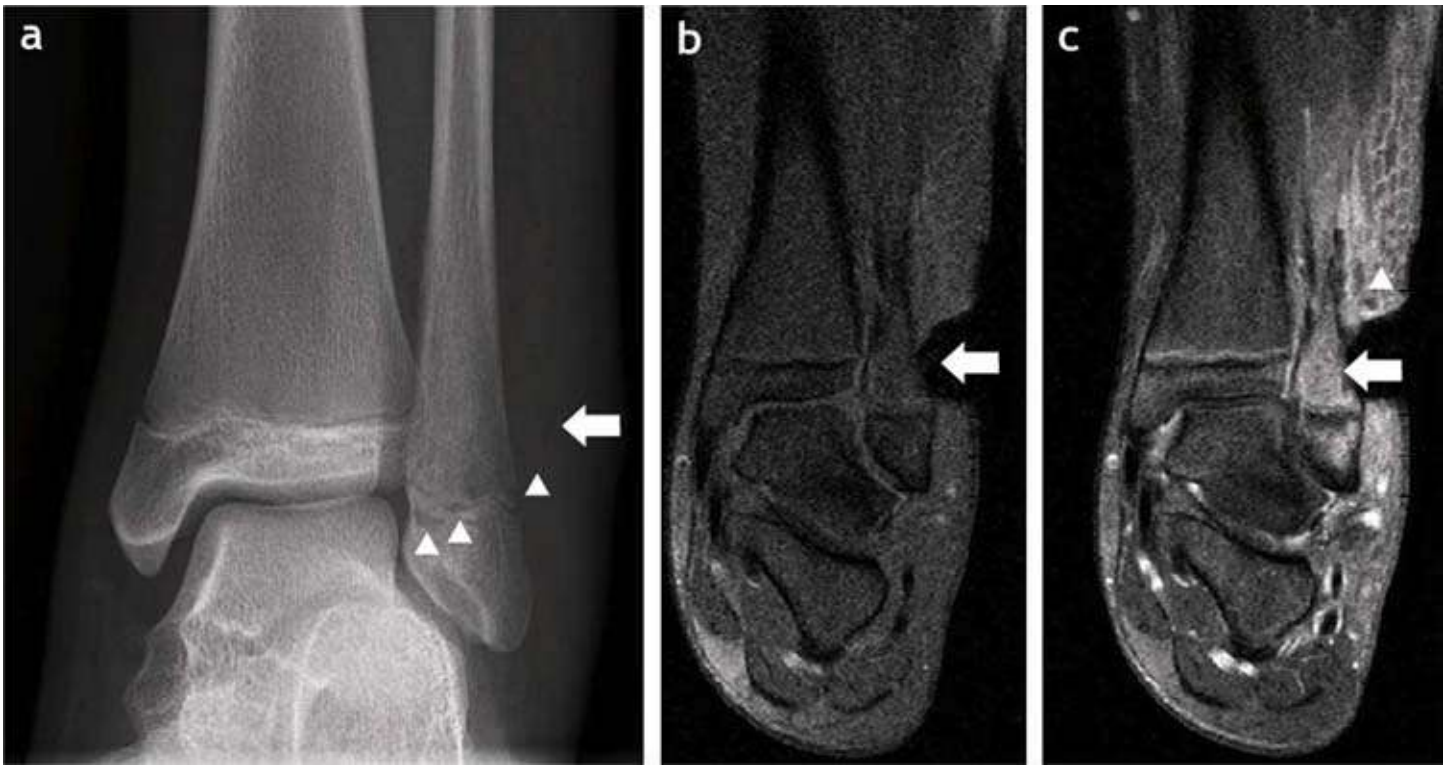

Figure 1:

a. Anterior posterior radiograph of the left ankle, demonstrating soft tissue swelling (arrow) and lytic bone lesions in the distal fibula (arrowheads).
b. Coronal T-1 weighted, fat suppressed magnetic resonance imaging (MRI) scan of the left ankle, showing a large skin and subcutaneous defect (arrow), where incision and drainage of the abscess was performed.
c. The same image after intravenous contrast (gadolinium), showing contrast enhancement in the distal fibula, corresponding with osteomyelitis (arrow). Abscess formation (arrowheads) and edema in the surrounding subcutaneous tissue can be seen.

While a multifocal presentation is common in neonates, where around 50% of the cases involve multiple bones[7], this is unusual in older children, occurring in 6-9% of those cases.[8,9] Multifocal osteomyelitis can also be caused by chronic recurrent multifocal osteomyelitis (CRMO), a rare disease that presents with sterile osteomyelitis with an unclear, presumably autoimmune etiology.[19]

We report an unusual case of multifocal AHOM in a 13-year old girl. The aim of this study is to emphasize that this diagnosis should be considered when a child presents with multifocal bone pain and/or painful joints, resembling a multifocal arthritis.

Case report

A 13 year-old girl was referred to our hospital with several painful joints since two days, accompanied by vomiting and fever. Physical examination showed a swollen right knee and a red, swollen proximal interphalangeal joint of the third digit of the left hand.

The patient was not severely ill with a non-febrile temperature. Blood tests showed a slight elevation of inflammatory and hematological markers: erythrocyte sedimentation rate (ESR) 24 mm/hour, C-reactive protein (CRP) 31 mg/l, serum ferritin 160µg/l, leukocyte count 12.6*109/l, neutrophil count 10.3*109/l.

The working diagnosis was reactive arthritis. The differential diagnosis consisted of juvenile idiopathic arthritis and septic arthritis, although low levels of inflammatory markers and a non-critically ill patient made these diagnoses unlikely. Diclofenac 50 mg was prescribed and the patient was sent home.

Over the next week, joint pain had not subsided and the right shoulder and right side of the pelvis were now involved. Furthermore, during the nighttime, the patient had developed a fever in excess of 40 degrees Celsius (104 degrees Fahrenheit), with a normal temperature during the day. Physical examination showed a swollen, warm and red area superior of the left lateral malleolus, as well as swollen digits 2 and 3 of the left hand. In addition, a painful area on the right iliac crest and slightly swollen knees were seen. Blood tests showed rising inflammatory markers (ESR 90 mm/hour, CRP 158 mg/l, serum ferritin 279 µg/l) and a rising leukocyte (15.2*109/l) and neutrophil count (11.6*109/l). Under suspicion of septic arthritis or osteomyelitis, the patient was admitted to the pediatric ward for further examination and treatment. A blood culture was done and the orthopedic surgeon performed a joint puncture of the ankle, which yielded clear liquid. The patient was treated with indomethacin and intravenous amoxicillin/clavulanic acid 2000/200mg three times daily. Three days after admittance, results of the joint puncture and blood culture were available. No bacterial growth was detected in the synovial fluid. The blood culture was positive for Staphylococcal aureus, after which antibiotics were switched to intravenous flucloxacillin 1000mg, four times daily.

In the meantime, her body temperature had dropped to normal levels; however an increase in pain and swelling of the left ankle was reported, with accompanying rising inflammatory markers (ESR 98 mm/hour, CRP 178 mg/l, neutrophil count 16.1*109/l and leukocyte count 18.7*109/l). Ultrasound of the left ankle showed a fluctuating, hypodense lesion, cranial to the lateral malleolus. Since an abscess was suspected, an incision was performed, after which a large amount of pus was drained. The periosteum had been damaged and underlying bone was visible, but still firm. The wound was thoroughly cleansed and was allowed to heal by second intention.

Further radiological evaluation was performed. Ultrasound of the pelvis showed a fluid collection lateral of the right anterior superior iliac spine (ASIS). Magnetic resonance imaging (MRI) scans of the pelvis showed bone edema in the right ASIS, and high signal intensity after intravenous contrast injection, corresponding with osteomyelitis. Pathological fluid pockets were seen in the soft tissue surrounding the affected bone, with capsular enhancement after contrast injection, corresponding with multiple micro-abscesses. MRI scans of the left ankle showed bone edema in the distal fibula and high signal intensity after intravenous contrast injection, corresponding with osteomyelitis. Pathological fluid pockets with capsular enhancement after contrast injection were also seen, pointing to micro-abscesses in the soft tissue surrounding the distal fibula. (Fig. 1)

Since multiple sites were affected, the presumptive diagnosis was multifocal acute hematogenous osteomyelitis.

Pain and limitation of range of motion gradually decreased over the next six days, with blood tests showing ever decreasing inflammatory markers (ESR 54 mm/hour and CRP 9 mg/l) and leukocyte and neutrophil numbers within the normal range. Three weeks after the onset of symptoms, the patient was discharged from our hospital with oral antibiotic therapy consisting of oral clindamycin 450 mg four times per day, which was to be continued for 4 weeks. At the outpatient clinic she showed complete recovery after 6 weeks.

Discussion

Diagnosis of multifocal AHOM can present a challenge because of the atypical presentation and similarity to septic arthritis, juvenile idiopathic arthritis and/or reactive arthritis. When AHOM presents in the pelvis, diagnosis can be especially difficult because pain is referred to the hip, thigh or abdomen because of the deep localization of the infection.[10] In one study, only 12 out of 82 patients suffering from pelvic AHOM were admitted with the correct diagnosis.[11]

Presenting clinical features of AHOM are pain, fever and in pelvic AHOM, limping. Laboratory findings include elevated inflammatory markers (ESR and CRP), which are found in over 90% of cases.[12] Peripheral white cell count is not a very reliable indicator of AHOM[1], however, white cell count should be performed, as leukemia is a differential diagnosis of AHOM. In this case, septic arthritis, juvenile idiopathic arthritis and reactive arthritis as well as leukemia were considered. Based on the not very ill patient and elevated inflammatory markers, the working diagnosis was reactive arthritis. Leukemia could be ruled out easily, based on the white cell count and differentiation, which is routinely performed in our hospital in similar cases.

Two important imaging modalities in the diagnosis of AHOM are bone scintigraphy and magnetic resonance imaging (MRI). These techniques are complementary. Hot spots on bone scintigraphy can guide high-resolution imaging modalities such as MRI, which have a limited field of view compared to a bone scan[10], but a higher sensitivity and specificity (93% and 96%).[13] Initial MRI is recommended in clearly localized disease, initial bone scintigraphy in diffuse, not clearly localized disease.[10] Since in this case, although disease was multifocal, symptoms were clearly localized, an indication for initial MRI scanning was present.

Bone or abscess aspiration to confirm the diagnosis is strongly recommended by experts. However, this is becoming increasingly controversial because of the high sensitivity and specificity of radiologic techniques to diagnose AHOM and S. aureus as causative pathogen in the majority of cases. Steer, et al., recommend incision and drainage when certain criteria are met, such as delayed presentation, an immunocompromised patient, abscess formation, underlying malignancy or delayed response to antibiotics.[1] In this case, abscess formation was a reason for incision and drainage of the ankle abscesses, which served as a combined interventional and diagnostic approach. Arthrocentesis of the ankle joint, performed under suspicion of septic arthritis yielded sterile synovial fluid.

A lesson to be learned from this is that AHOM close to a joint can clinically mimic septic arthritis. Therefore, when arthrocentesis is performed in case of a clinically suspected septic arthritis and sterile fluid is obtained, the clinician should consider osteomyelitis close to the joint.

Antibiotic therapy is initially based on the most likely pathogen causing AHOM in children, which is S. aureus. Single β-lactamase-resistant penicillins such as flucloxacillin have shown their effectiveness against methicillin-susceptible staphylococci and streptococci.[14] After identification of the organism, antimicrobial therapy can be changed according to bacterial susceptibility. When blood/bone culture turns out negative, empirical antibiotic therapy can be continued, as long as clinical response is observed. Duration of antibiotic therapy in AHOM differs from that in adults. Adults are usually treated with prolonged intravenously therapy (mostly six weeks). Children can be treated for shorter courses and with parenteral-oral sequential therapy[1], in which intravenous administration is followed by oral continuation of antibiotic therapy. The efficacy and safety of this regimen has been proven in numerous studies, some of which measuring bone concentrations of oral antimicrobials.[15,16] The total duration of therapy and timing of the switch to oral therapy differs greatly between studies, but generally, intravenous therapy for 3-7 days and further continuation of oral therapy for 3-4 weeks is most common[17,18], although this has never been thoroughly tested in a randomized controlled trial.[19]

Conclusion

In our opinion, two important lessons can be learned from the aforementioned case. Firstly, alternative diagnoses such as multifocal AHOM should be considered when patients present with atypical symptoms, including bone or joint pain and fever. Secondly, when arthrocentesis is performed under suspicion of septic arthritis and sterile synovial fluid is obtained, this can very well mean AHOM close to the apparently symptomatic joint. This case underscores the importance of careful consideration of different etiological entities and multidisciplinary diagnosis and treatment when a patient presents with atypical symptoms and signs.

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