Case Report
Volume 11:12, 2021

Clinical Case Reports

ISSN: 2165-7920 Open Access

Bone Marrow Edema Syndrome in the Ankle: Case Reports and Literature Review

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Abstract

Bone Marrow Edema Syndrome (BMES) is a clinical syndrome of unknown etiology characterized by the acute onset of pain gradually worsening over several weeks to months. Radiographic changes occur, but laboratory studies are generally unremarkable. It can primarily affect the foot and ankle and should be considered in the differential diagnosis of patients with acute foot and ankle pain, particularly in middle age men, and women in the third trimester of pregnancy. I here present 2 case reports of BMES of the ankle. Appropriate conservative treatment resulted in resolution of symptoms. Bone marrow edema syndrome can present a diagnostic challenge, but awareness of the typical presenting features and investigation findings can makes the diagnosis more accessible.

Keywords: Bone marrow edema • Bone marrow edema syndrome • Transient osteoporosis

Introduction

Bone Marrow Edema Syndrome (BMES) is a relatively recent term used to describe a spectrum of conditions characterized by pain and increased interstitial fluid within the marrow of a bone, with detection of Bone Marrow Edema (BME) on Magnetic Resonance Imaging (MRI) as an isolated finding without apparent cause [1,2]. The etiology and pathogenesis of this condition are unknown, and it most commonly affects middle-aged men and women in the third trimester of pregnancy [3]. BMES most commonly affect the hip, followed by the knee, ankle and foot, although the available data has been generally retrieved from relatively small series or from case reports rather than epidemiological studies [4]. The patient presents with joint pain which is usually of spontaneous onset and exacerbated by weight bearing if the lower limbs are involved [2,4]. Usually, only one joint is affected, but more than one joint may be involved consecutively; a condition known as Transient Migratory Osteoporosis (TMO) [2]. BMES of the foot and ankle is an infrequent and often under- recognized disorder [5]. The correct diagnosis in the foot and ankle often is delayed because of the low prevalence and nonspecific signs [6]. I here report 2 cases of BMES of the ankle and foot, which illustrates the importance of recognizing the typical clinical and radiological features of this rare condition, thereby allowing an appropriate management.

Case Report

Case 1

A 23 year-old woman presented to our rheumatology department with complaint of right ankle pain for 5 months duration. Her condition started when she was 7 months pregnant. The symptoms seemed to appear spontaneously, there were no identifiable factors which might have precipitated the pain. There was no clear history of antecedent trauma. The pain began gradually and became more severe and diffuses over a period of 2 to 3 weeks. The pain gets aggravated on weight bearing and relieved with rest. She walked with a limp because of the pain, and most of the time needed assistance. Morning stiffness was not present. The patient reported no fever, weight loss, diarrhoea, or skin rash. The patient past medical history was unremarkable, and her family history was not contributory.

On physical examination there was mild swelling, involving the ankle and mid foot. The synovial membrane was not thickened. The right ankle was mildly tender on palpation, but the skin was not cold, clammy, moist, cyanotic, or atrophic. Ankle Active and passive range of motion were normal with only slight pain at the extreme. Laboratory findings including full blood count, erythrocyte sedimentation rate, C-reactive protein, liver function tests, creatinine, electrolytes, thyroid function test and protein electrophoresis were normal. Tests for anti-nuclear body, rheumatoid factor were negative.

Radiographs of the right ankle and foot showed gross patchy osteopenia with no clear evidence of joint destruction or fracture. The decrease in bone density was most marked in the talus and tarsus bones. The calcaneus, distal end of tibia and fibula showed lesser degree of involvement (Figure 1). MRI of the left ankle showed Mild ankle and subtalar joint effusions, with high signal intensity on T2-weighted images and low signal intensity on T1-weighted

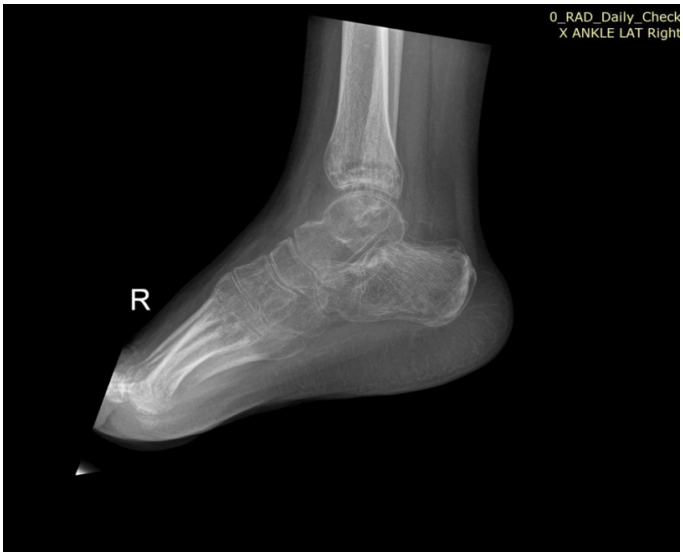

Figure 1. Radiographs of the right ankle and foot (lateral view) shows gross patchy osteopenia most marked in the talus and tarsus bones. The calcaneus, distal end of tibia and fibula showed lesser degree of involvement. No clear evidence of joint destruction or fracture.

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Received 06 December 2021; **Accepted** 15 December 2021; **Published** 22 December 2021

images are seen mainly involving the talus, and to a lesser extent the distal tibia and fibula, calcaneus and other tarsus bones (Figure 2).

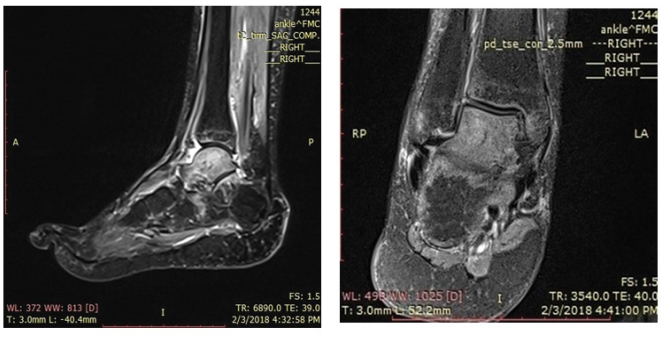
The patient was treated with protected partial weight-bearing, prednisolone 20 mg daily which is tapered over 1 month, risedronate 35 mg once weekly for 3 months, and physiotherapy. The physiotherapy included active and passive range of motion exercises for 4 weeks. After 4 weeks, the patient came again to our rheumatology clinic with marked relief in her pain. She was then walking with one stick. Four weeks later her symptoms considerably resolved, and she could walk with minimum pain and no limp. The patient recovered fully after 3 months of follow-up, and she regained her normal gait.

Case 2

A 54 year-old man presented to our rheumatology department with gradually increasing pain in his right ankle for 1 week duration. There was no any history of antecedent trauma. The pain began acutely and became more severe and diffuses over a period of one week. The pain gets aggravated on weight bearing and relieved with rest. He walked with a limp because of the pain. Morning stiffness was not present. The patient reported no fever, weight loss, diarrhea, or skin rash. He also had a history of right knee pain 10 months back which was insidious in onset and gradual in progression, aggravated by weight bearing and relieved by rest. His pain did not improve with treatment including moderation of activity, and anti-inflammatory medications. Magnetic Resonance Imaging (MRI) of knee was done 2 months later, which showed high signal intensity in the medial femoral condyle on T2-weighted images and low signal intensity on T1W images (Figure 3). His pain resolved spontaneously over 7 months. The family history was not contributory.

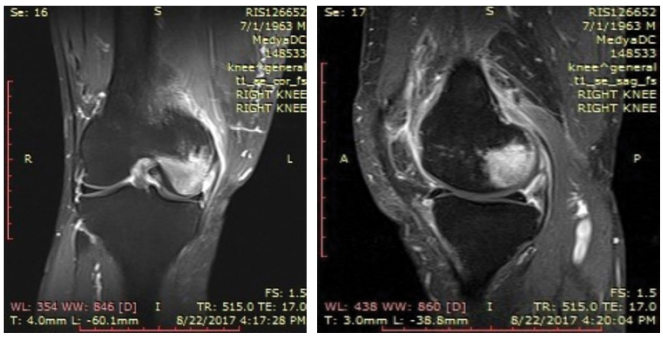
On physical examination there was mild swelling, involving the right ankle with normal overlying skin. The right ankle and lower tibia were mildly tender on palpation. Ankle Active and passive range of motion were normal with only slight pain at the extremes. Peripheral pulses were intact, with unremarkable neurological evaluation. Laboratory findings including full blood count, erythrocyte sedimentation rate-reactive protein, liver function tests, creatinine, electrolytes, thyroid function test and protein electrophoresis were normal. Tests for anti-nuclear body, rheumatoid factor were negative. Plain radiographs of the ankle were unremarkable.

MRI of the right ankle showed mild joint effusion with high signal intensity on T2-weighted images and low signal intensity on T1-weighted images involving the superior aspect of the talus as well as the distal tibia (Figure 4). The articular surfaces were intact, with no fracture line, osteochondral lesions or osteonecrosis. The findings were consistent with BME syndrome. The patient was treated with protected weight bearing, calcitonin nasal spray, prednisolone 20 mg daily which is tapered over 1 month, risedronate 35 mg once weekly for 3 months, and physiotherapy. The patient recovered fully after 5 months of follow-up, and he regained his normal gait.



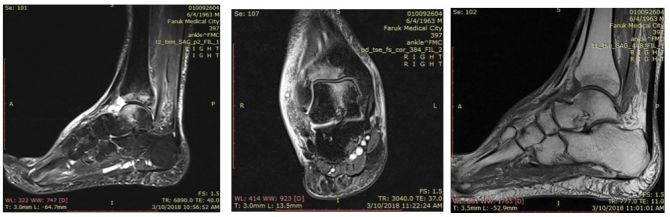
A B

Figure 2. MRI of the left ankle shows mild ankle and joint effusions, with high signal intensity on T2-weighted images (A and B) seen mainly involving the talus, and to a lesser extent the distal tibia and fibula, calcaneus and other tarsus bones.



A B

Figure 3. MRI of the right knee shows high signal intensity in the medial femoral condyle on coronal (A) and sagittal (B) T2-weighted images.



A B C

Figure 4. MRI of the right ankle shows mild joint effusion with high signal intensity on T2-weighted images (A,B) and low signal intensity on T1-weighted images (C) involving the superior aspect of the talus as well as the distal tibia. No evidence of fracture line, osteochondral lesions or osteonecrosis.

Discussion

The term BMES was used to describes a clinico-radiological entity in which transient non-specific subacute or chronic joint pain is associated with characteristic MR appearances in the absence of identifiable underlying cause [7]. Different terminologies have been used in the literature to describe BMES reflecting changes in the perception of the condition with time and by different specialties within medicine. The term transient osteoporosis was first used to describe a self-limited condition characterized by pain and regional loss of bone mineralization followed by spontaneous recovery [8]. The term regional migratory osteoporosis was used later to describe a recurrent and migratory pattern of transient osteoporosis [9]. The term bone marrow oedema was first used by Wilson AJ, et al. [10] the authors described MRI characteristics and clinical findings of transient osteoporosis of the hip and the knee and concluded that the findings represent edema in the marrow. They found no evidence of osteoporosis on examining histologic specimens, so they suggested instead the term transient bone marrow edema. The term BMES was suggested by Hoffman S, et al. [11].

The etiology and pathogenesis of BMES remains unclear. Commonly suggested explanations include viral infection, nerve compression syndromes, reflex sympathetic dystrophy, and an increased intramedullary pressure due to an altered venous outflow. Others have suggested “regional accelerated phenomenon” as a possible mechanism, in which bone tissue micro damage and consequent micro fracture may be the noxious stimuli that trigger this acceleration phenomenon, in which the rate of bone modelling and remodelling in local areas may be increased up to ten times [12].

It is important to stress that BMES should only be diagnosed once other known causes of BME have been excluded. These include posttraumatic injury, stress fractures, degenerative and inflammatory joint disorders, neuropathic joint disease, complex regional pain syndrome, infection, and malignancy [1,12].

Clinically, the patient usually presents with joint pain of spontaneous onset. The pain has variable intensity, sudden or insidious onset, which is exacerbated by weight bearing, improves with rest and may deteriorate quickly

and progressively until it causes severe functional disability which requires hospitalization. A key clinical feature is that the symptoms and disability are out of proportion with clinical findings, although it is sometimes accompanied by inflammation, with soft tissue edema and a slight joint effusion [4]. It is characterized radiographically by periarticular demineralization of one bone 4-8 weeks after the onset of pain. There are no bony erosions, narrowing of the joint space or subchondral fractures. Remineralisation usually takes place within 2 years. MRI is the diagnostic method of choice for BMES. Forty-eight hours after the onset of the symptoms the characteristic signal pattern of the bone marrow edema can be observed. In some cases there is also a joint effusion [13]. MRI reveals low-signal intensity on T1-w images and high signal intensity on STIR or fat-suppressed T2-w images [12].

Most series emphasized the self-limiting nature of symptoms; the time taken for the improvement of symptoms and changes in MRI is between six and 12 months [4]. However, some authors have reported cases in which there is lack of complete remission beyond 12 months [4,14].

The treatment of BMES is usually conservative, with rest, analgesics, anti-inflammatory drugs, unloading and physical therapy. Several drugs have been used in this condition including glucocorticoids, bisphosphonates, and iloprost,a prostaglandin analogue. The most recommended bisphosphonates drugs are alendronic acid, pamidronic acid, ibandronic acid and zoledronic acid. Bisphosphonates reduce bone turnover by inhibiting osteoclastic activity and reducing bone resorption, and can be used orally or intravenously. Although their mechanism of action in BMES is not entirely clear, bisphosphonates may have an anti-inflammatory effect. The addition of bisphosphonates may hasten the recovery [1,15].

Conclusion

BMES of the foot and ankle is a clinical disorder that should be rule out in all pregnant women and middle age men who present with a clinical history of prolonged foot and ankle pain of unknown etiology and without prior trauma. The pain is usually mechanical, progressively disabling and disproportionate to the clinical signs. An early diagnosis with the use of MRI is important. Also, an early treatment is essential for a quick recovery from this condition and to prevent further unnecessary costly and invasive investigations.

Conflict of Interest

The authors declare no potential conflict of interests

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How to cite this article: Younis, Ali Abdulrahman. "Bone Marrow Edema Syndrome in the Ankle: Case Reports and Literature Review." Clin Case Rep 11(2021): 1482.