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Case Report

A Case of Transient Migratory Osteoporosis of the Right Knee and Ankle in a 46-Year-Old Male

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Abstract

Transient Osteoporosis (TO) is an uncommon skeletal condition typified by pain in weight bearing joints and bone marrow oedema on MRI, both of which resolve spontaneously. TO most frequently affects middle-aged men and women in the third trimester of pregnancy, with the hip being most commonly affected. There is usually no preceding history of trauma or insult, and the symptoms generally worsen overtime and are exacerbated on weight bearing or physical activity. Resolution may take up to 6-9 months. The etiology of TO remains largely unclear but several theories have attempted to explain it. Strenuous activity, smoking and pregnancy, are a few of the risk factors quoted in the literature. The gold-standard diagnostic tool is MRI as other modalities often show no abnormalities. The main differential diagnosis for TO is avascular necrosis (AVN); it is crucial to distinguish between the two as AVN is progressive and destructive. Currently, the mainstay of treatment of TO revolves around conservative management although other innovative options are being studied such as core decompression and hyperbaric oxygen therapy.

Keywords: *Transient Osteoporosis; Primary Bone Marrow Oedema Syndrome; Regional Migratory Osteoporosis*

Abbreviations

MRI: Magnetic Resonance Imaging; DEXA: Dual-Energy X-Ray Absorptiometry; TO: Transient Osteoporosis; BME: Bone Marrow Oedema; PBMES: Primary Bone Marrow Oedema Syndrome; HCP: Health Care Professional; AVN: Avascular Necrosis; NSAIDs: Non-Steroidal Anti-Inflammatories Drugs; HBOT: Hyperbaric Oxygen Therapy

Introduction

Transient osteoporosis is a self-limiting disease, causing pain in weight bearing bones and is characterized by bone marrow oedema on MRI. Typically presents in middle aged men as pain in a weight bearing joint which is worse on exercise. The main stay of treatment is conservative however, it is important to distinguish it from avascular necrosis as these may present similarly but are managed differently.

Case Report

A 46-year-old gentleman who was otherwise healthy was seen at the orthopaedic clinic with a history of right sided knee pain. The pain was over both the medial and lateral aspects of the right knee, had started a few weeks earlier and gradually worsened. The patient also reported swelling of the same knee and worsening pain on high impact activities. This had been his first such episode, with no similar episodes in any other joints in the past. There was no history of preceding trauma, intervention, or surgical procedure.

Physical examination revealed moderate right knee swelling with underlying effusion and painful hyperflexion. There were no signs of collateral ligament pathology. Anterior and posterior drawer tests were both negative suggesting both cruciate ligaments were intact.

The patient had some routine blood tests taken, including an autoimmune screen. Of note, he had a marginally low phosphate and vitamin D. All other blood tests were unremarkable, and the autoimmune screen resulted negative.

An MRI of the knee was performed and noted diffuse marrow oedema-like signal change throughout the medial weight-bearing compartment, especially the femoral aspect, extending laterally. Subtle subarticular fracture lines were appreciated at the central and posterior thirds of the medial femoral condyle as well as the inferior third of the lateral femoral condyle. There was also a significant knee joint effusion. Both menisci, as well as other surrounding structures were intact. The overall appearance was very suggestive of transient osteoporosis at the right knee level.

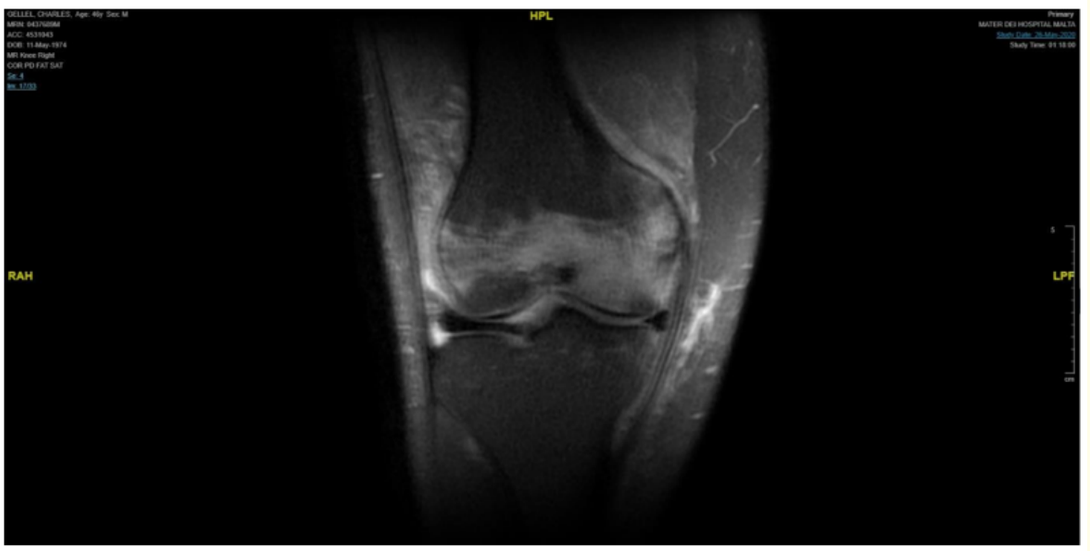

Figure 1: *Coronal section from first MRI of the knee that was performed highlighting bone marrow oedema namely of both the medial and lateral femoral condyles.*

The patient was advised limited weight-bearing and was prescribed calcium supplements. He was booked for a repeat scan in 3 - 6 months' time to assess for resolution. The follow up MRI showed incomplete resolution of the previously documented abnormalities present on both medial weightbearing surfaces. Interval resolution of the changes in the anterior half of the lateral femoral condyle was noted, however, more extensive changes developed throughout the posterior half of the lateral femoral condyle, with resultant diffuse marrow oedema-like signal change and linear subarticular sclerosis. No articular surface collapse or bony fragmentation was reported and there were no other significant findings. These findings were again suggestive of transient osteoporosis but screening for any possible underlying avascular necrosis predisposing factors was advised.

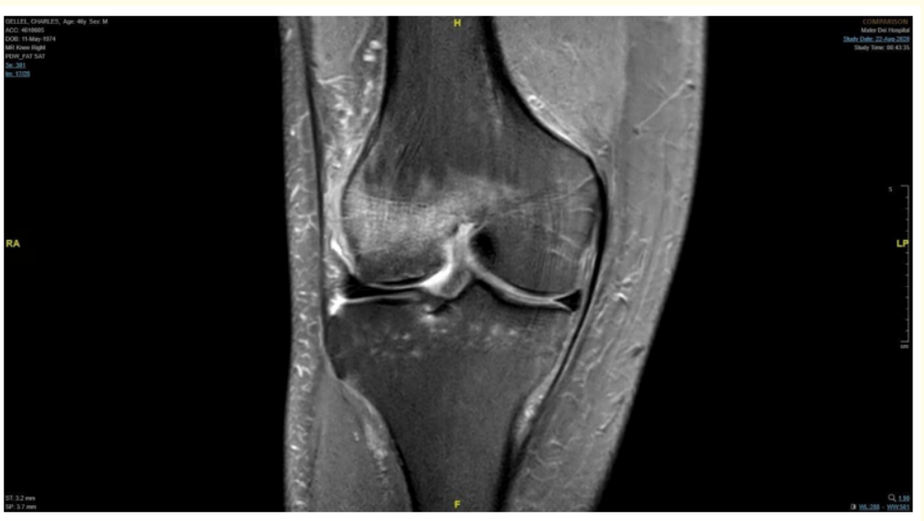

Figure 2: *Coronal section from second MRI of the knee that was performed highlighting persisting bone marrow oedema, this time namely of the medial compartment.*

The patient subsequently developed a sudden, atraumatic swelling of the right ankle. This was also investigated with MRI which showed significant bone marrow oedema-like signal intensity affecting multiple bones, including the talar head, with evidence of a subtle subchondral insufficiency fracture at its articulation with the navicular bone. Similar changes were noted at the intermediate cuneiform and the second metatarsal. Other structures were unremarkable. These findings affirmed a diagnosis of bone marrow oedema syndrome, specifically transient regional migratory osteoporosis (given previous similar MR findings in the right knee). Follow-up imaging and correlation with vitamin D levels and dual-energy x-ray absorptiometry (DEXA) scans and/or radiographs was recommended.

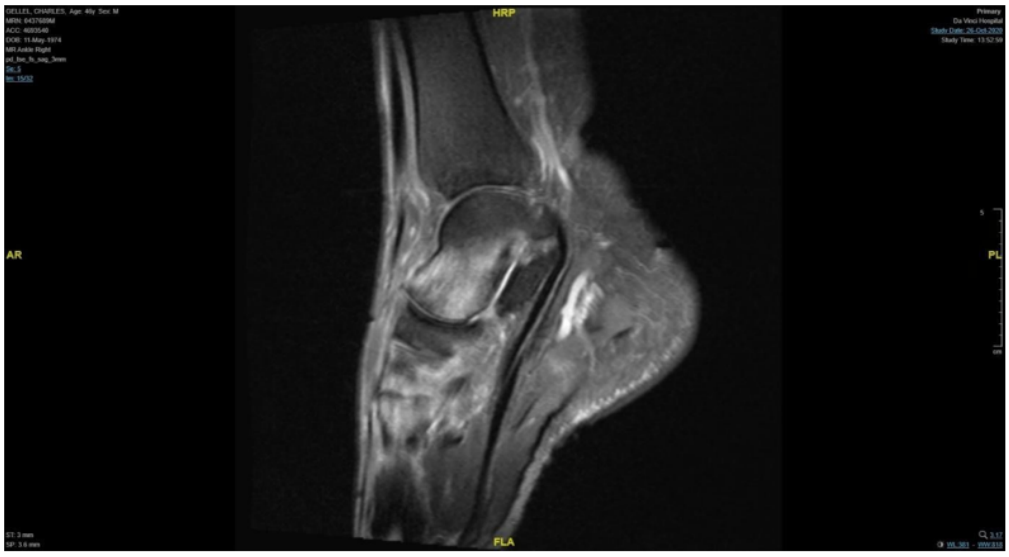

Figure 3: *Sagittal section from MRI of the ankle showing bone marrow oedema of the talar head.*

Discussion

Transient Osteoporosis (TO) is a rare, self-limiting skeletal condition which is characterized by pain in a weight bearing joint with no clear etiology, bone marrow oedema (BME) on MRI and spontaneous resolution within a few months [1-3]. The clinical presentation may be variable and non- specific and thus it is also known as Primary Bone Marrow Oedema Syndrome (PBMES), Regional Migratory Osteoporosis, as well as several others [4-6]. It was first described by Curtiss and Kincaid in 1959 where they reported on three cases of TO of the hip in pregnant women [2].

TO most commonly affects middle-aged men and women in the third trimester of pregnancy with a ratio of 3 to 1 when comparing the incidence in men and women [7]. It primarily affects weight bearing joints in the order of the hip, knee, foot, and ankle. Occasionally, it may occur bilaterally [8]. The disease has been known to recur, affecting the same or other joints and, in these circumstances, it has been termed regional migratory osteoporosis.

The classical presentation of TO is spontaneous pain in a weight bearing joint with no preceding history of trauma or insult. The symptoms generally worsen overtime and are exacerbated on weight bearing or physical activity. A three-stage process is consistently described in the literature; an initial stage with onset of pain and loss of function, followed by a second stage characterised by maximal pain and, finally, a third stage during which there is resolution of symptoms [5,9]. Spontaneous resolution is typical of TO. Different durations of effect are quoted but most range between six to nine months, after which there is both clinical and radiological resolution [2,6,10].

Several theories have been proposed suggesting the likely etiology of TO, however strong evidence is lacking and thus the etiology remains unclear. These theories commonly centre around the concept of venous obstruction and stasis within the affected bone, with thromboembolism, coagulopathic factors and high level of lipoprotein and plasminogen activator inhibitor all being implicated as a possible causative agent [5,9,11,12]. Trauma, sudden increase in activity, pregnancy, smoking, alcohol misuse, steroids, illicit drugs, previous episode of TO, osteogenesis imperfecta, hypothyroidism and vitamin D deficiency have all been quoted as possible risk factors for developing TO [3]. A recent study by Bashaireh., *et al*. demonstrated that occupation, namely that of a health care professional (HCP), may be an independent risk factor for developing TO as 9 out of a cohort of 15 patients that were studies were HCPs [12].

TO is a diagnosis of exclusion and several investigations need to be performed before committing to the diagnosis. Usually, biochemical analysis and plain radiographs are unremarkable, especially early on. Changes on plain radiographs are usually noted around 4 weeks after symptom onset and include widespread osteoporosis of the involved bone [12]. CT scans may be performed to rule out other causes of pain but the investigating of choice when investigating for TO is magnetic resonance imaging (MRI). The classic findings are universally documented in the literature [2,5,10,12,13]:

- An area of homogenous decreased signal intensity on T1-weighted images.
- A corresponding area of homogenous increased signal intensity on T2-weighted images.
- No masses, no focal areas of osteonecrosis, intact articular surface and no double-line sign.

An important distinction must be made between TO and avascular necrosis (AVN). The two may have similar presentations, with pain in a weight bearing joint being typical, and also have similar risk factors [10]. McCarthyetal demonstrated distinctive histological features in samples from patients with TO namely, marrow oedema, secondary bone formation and resorption of bone by osteoclasts. Crucially, no necrosis was visualised, suggesting the distinction from AVN. It is important to make this distinction as the outcome and prognosis of the two varies significantly. While TO resolves spontaneously within a few months with just conservative management, AVN is progressive, more destructive and requires surgical intervention [14].

The mainstay of treatment for TO is conservative management, namely rest and offloading of the affected joint as well as analgesia using drugs such as non-steroidal anti-inflammatories (NSAIDS). Several other remedies have been documented and investigated including oral and intravenous bisphosphonates, prostacyclin derivatives, hyperbaric oxygen therapy and surgical management with core decompression [15,16]. Strong evidence regarding the optimal therapy for TO is lacking. A systematic review by Ververidis., *et al*. concluded that the use of bisphosphonates to achieve pain relief and early regression was controversial and no recommendations could be made as to their use [15]. A prospective, randomized trial by Capone., *et al*. Compared TO patients receiving conservative treatment (protective weight-bearing and NSAIDs) to those receiving conservative treatment with hyperbaric oxygen therapy (HBOT). The study showed that those patients receiving HBOT had faster recovery times on MRI as well as faster resolution of symptoms when compared to those who did not receive HBOT. They concluded that the use of HBOT may be used as an alternative in patients with non-resolving TO, highlighting the disadvantage that several sessions were required and that data on the treatment was still lacking [17]. Data on the role of core decompression in the management of TO is also lacking and is limited to a handful of case reports. The theory is that core decompression partially relieves the intramedullary pressure and allows for decompression of the surrounding bone marrow [18].

Conclusion

TO is a rare condition mainly affecting middle-aged men and women in the third trimester of pregnancy. It is characterized by pain in a weight bearing joint, usually the hip, with no specific cause and bone marrow oedema on MRI. Typically, there is both clinical and

radiological resolution within 6 - 12 months. MRI findings classically demonstrate decreased signal intensity on T1-weighted images and increased signal intensityonT2-weighted images. The optimal management of TO remains unclear due to a lack of strong, evidence-based research. However, most authors endorse conservative methods, with offloading of the affected joint and simple analgesia. As previously stated, there is a lack of evidence regarding the etiology, pathogenesis and optimal management of this rare disease so further studies are required to better understand it and recommend optimal management.

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