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Case Report

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Transient Osteoporosis of the Knees, Ankles and Feet: Atypical Case in a Twin Pregnancy

Tatiana Elizabeth Carranco-Medina[a, b, c], Ismael Calero-Paniagua[a, b], Cristina Hidalgo-Calleja[a, b], Maria Dolores Sanchez-Gonzalez[a, b], Carlos Alberto Montilla-Morales[a, b], Alba Quesada-Moreno[a, b], Ricardo Usategui-Martin[b], Laura Perez-Garrido[a, b], Susana Gomez-Castro[a, b], Olga Martinez-Gonzalez[a, b], Javier Del Pino-Montes[a, b]

Abstract

Transient osteoporosis (TO) is a rare entity with unknown origin which is, in most cases, self-limiting. The most common location is the hip, which can sometimes be bilateral, followed by the knee, ankle and foot. The multifocal, bilateral and simultaneous location of knees, ankles and feet during pregnancy is extremely rare. We present the case of a woman, 25 weeks pregnant with twins, who was admitted as an emergency with pain and functional disability of her lower limbs. She required hospitalization and was diagnosed with transient osteoporosis of pregnancy (TOP), with clinical and radiological resolution after 12 months and treatment with intravenous bisphosphonate.

Keywords: Transient osteoporosis; Transient bone marrow edema syndrome; Pregnancy; Twins

Introduction

Transient osteoporosis (TO) is a rare disease which may affect middle-aged men and pregnant women in their third trimester or after delivery [1, 2]. Some of the terms commonly used to describe this medical entity are transient bone marrow edema syndrome, transitory bone demineralization, regional migratory osteoporosis, reflex sympathetic dystrophy, etc. [3]. It presents with spontaneous joint pain and functional loss without a previous traumatic record. Simple X-rays and bone scintigraphy are useful to confirm a diagnosis, and magnetic resonance imaging (MRI) is the most sensitive technique [3, 4].

The incidence and prevalence of this condition is unknown given the lack of published series, and there are only some isolated cases reported in pregnant women. Curtiss and Kincaid published the first cases in 1959 [5] and, since them, several cases have been reported with the hip joint as the most common location, followed by the knee, ankle and foot. There are few published articles with a simultaneous involvement of both joints of the lower limbs. Our case presents a woman pregnant with twins with simultaneous bilateral involvement of knees, ankles and feet.

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[a]Hospital Universitario de Salamanca, Rheumatology Department, Spain
[b]Salamanca Institute for Biomedical Research, IBSAL, Spain
[c]Corresponding Author: Tatiana Elizabeth Carranco-Medina, Hospital Universitario de Salamanca, Rheumatology Department, Spain.
Email: tatyeli21@hotmail.com

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Case Report

The patient is a 36-year-old woman, 25 weeks pregnant with twins, who was admitted as an emergency with intense pain on the knees and ankles, without a previous record of trauma, associated with functional disability for standing and walking, which required hospitalization. The physical examination revealed intense pain on palpation and on mobilization of knees and ankles. No joint effusion was observed and the meniscal maneuvers were negative. Muscle strength and sensitivity were normal. The knee ultrasound was also normal, and the MRI of the right knee showed a slight joint effusion with bone edema on the subchondral region of the patella and the internal femoral condyle, with a small image of a microfracture (Fig. 1). The analysis revealed hypercalcemia of 11.3 g/dL, vitamin D of 15 mg/dL, and hypercalciuria of 665 mg/day. Complementary tests were performed and a malignant process was ruled out.

After a diagnosis of osteoporosis of pregnancy (TOP), treatment was started with analgesics, topical capsaicin and rest. The patient did not report a clinical improvement and she presented with class IV functional capacity. A programmed cesarean section was performed in week 35 of pregnancy. In the postoperative period, X-rays of knees, ankles and feet were obtained (Fig. 2), as well as a bone scintigraphy (Fig. 3). Given the findings and the clinical symptoms, which were resistant to the usual treatment, a 5 mg dose of intravenous zoledronic acid was administered, associated with calcium, vitamin D and rehabilitation.

The control bone scintigraphy which was performed 3 months later detected a lower enhancement of technetium 99 tracer (Fig. 4), as well as normal figures for Ca and vitamin D in the analytical tests.

After 8 months, the patient showed a clinical and radio-

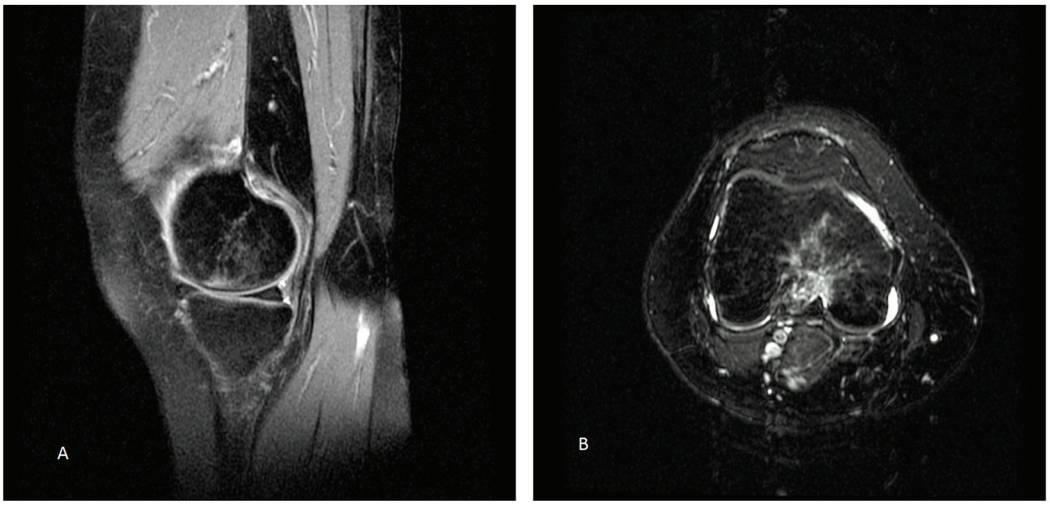

Figure 1. MRI in T2. Enhanced signal on the internal femoral condyle: (A) sagittal section and (B) axial section.

logical improvement (Fig. 5), with functional class I.

Discussion

TO is a rare entity, but it is cited with increased frequency, and it is probably underdiagnosed. Up to date, there are no adequate epidemiological studies, and its incidence is estimated based on the few clinical cases and series which have been published. It is located on the load-bearing joints of the lower limbs: hip (70%), knee, ankle and foot [6]. An involvement of different areas of the same joint is also possible, as well as migration from one joint to another, which is described as regional migratory osteoporosis. From 10 to 40% of patients develop this condition in several joints, either simultaneously or successively, on the same limb or on the contralateral limb. In its most common presentation, it spreads from the proximal to the distal end in the same limb [6-9]. A bilateral simultaneous involvement in both hips is exclusive of pregnant women [9, 10]. Its pathogenesis is unclear and it is suggestive of a multifactorial origin: microtraumatisms, trabecular microfractures caused by a reduction of bone mass during pregnancy, neurovascular dysfunction, joint overload caused by weight gain and hormonal changes typical of pregnancy, resulting in ischemic tissue and bone edema, which are characteristic of this entity [6, 10].

Clinically, it presents with spontaneous pain without a previous traumatic or infectious record, with variable intensity, sudden or insidious onset, which is exacerbated by loads, improves with rest and may deteriorate quickly and progressively until it causes severe functional disability which requires hospitalization. One of its main clinical characteristics is that symptoms and disability are not correlated with clinical findings, although it is sometimes accompanied by inflammation, with soft tissue edema and a slight joint effusion. Fatigue fractures are a rare complication [8].

The analytical study does not generally reveal alterations,

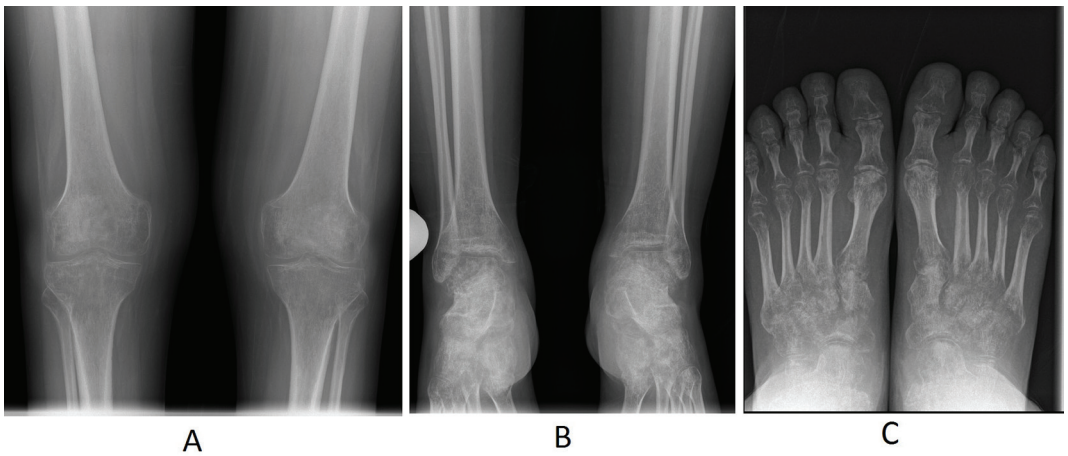

Figure 2. AP simple X-ray: (A) diffuse osteopenia on the distal femoral metaphysis and epiphysis and on the proximal tibia and fibula; (B) patchy osteopenia on the distal metaphysis and epiphysis of tibia and fibula and on tarsi; (C) patchy osteopenia on tarsal bones, head and base of metatarsal bones and phalanges.

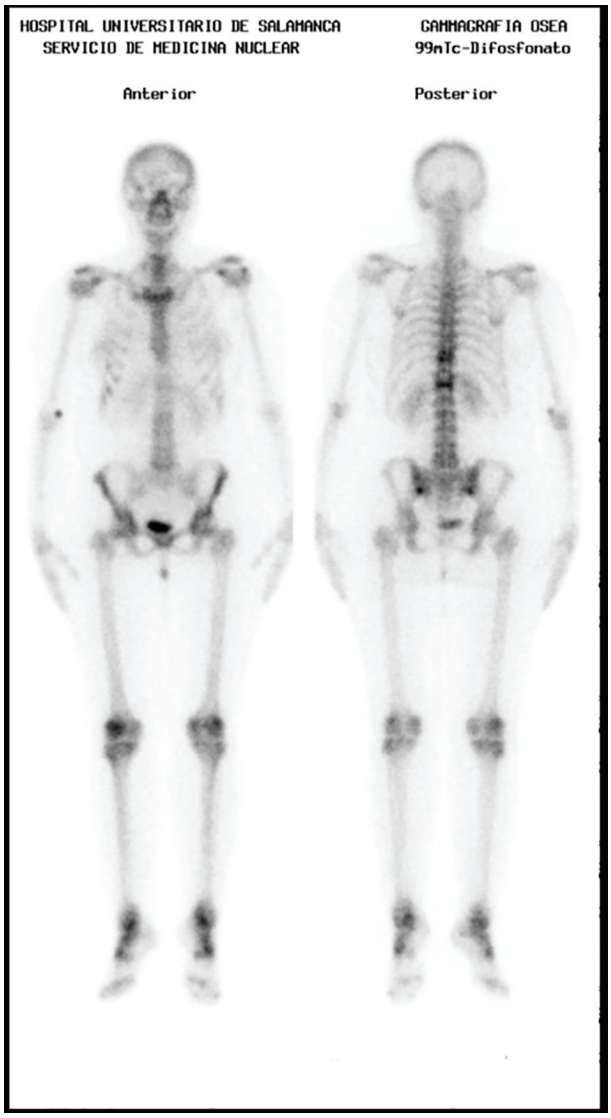

Figure 3. Bone scintigraphy with technetium 99: enhancement of the tracer on the knees, ankles, tarsal and metatarsal bones.

except for those typical of pregnancy. In our case, hypercalcemia might be due to the large extension of the bone involvement and the immobility caused by the severity of the functional disability.

In its first stages, X-rays are not able to detect alterations in bone mineralization although more advanced stages may reveal diffuse osteopenia with preservation of joint spaces. At an early stage, scintigraphy is a more sensitive technique and it shows marked and diffuse hyper-enhancement of the isotope. MRI is an essential tool in diagnosis; it shows bone edema in the affected area, it makes it possible to obtain a differential diagnosis with other processes, mainly infectious and cancerous, and it is also extremely useful during the monitoring stage [6, 7, 10, 11].

Most series highlight the self-limiting nature of symptoms over 6 - 12 months. However, in some patients, this period may be longer, depending on the extension and severity of the form

Figure 4. Bone scintigraphy with technetium 99: decreased enhancement of the tracer.

of presentation as well as on the treatment [6, 11].

The treatment of choice is conservative, with rest, analgesics, anti-inflammatory drugs, unloading and physical therapy. Several drugs have been used in these cases. However, evidence is based on the experience of published clinical cases and case series. Some of the drugs which have been used are bisphosphonates, glucocorticoids, prostaglandins, and calcium antagonists. In the group of bisphosphonates, the most recommended drugs are alendronic acid, pamidronic acid, ibandronic acid and zoledronic acid [10, 12].

Lopez Sagasta et al described the case of a 46-year-old woman pregnant with twins in her third trimester who developed a unilateral transient bone edema of the knee with spontaneous resolution after 8 - 9 months [13]. Our case is atypical due to the simultaneous and bilateral involvement of several joints of the lower limbs. We also believe that the twin pregnancy contributed to this aggressive form of presentation and that the adequate postpartum treatment with intravenous bisphosphonates improved the evolution of this condition.

It is necessary to rule out the presence of TO in all pregnant women who, in their third trimester, present with permanent pain of one joint which is mechanical, progressively disabling and disproportionate to the clinical signs.

Figure 5. AP simple X-ray: (A) disappearance of patchy osteopenia of the knees; (B) marked reduction of diffuse osteopenia on tarsal and metatarsal bones and on phalanges.

An early diagnosis with the use of scintigraphy and MRI, whenever the circumstances allow it, is important. Also, an early treatment is essential for a quick recovery from the process and to prevent complications such as fractures.

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