Bilateral transient osteoporosis of the knees during pregnancy. A case report and review of the literature

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Introduction

Transient osteoporosis (TO) is a rare self-limited condition and there have been less than 200 cases of this disease reported in the literature. In 1959, Curtish and Kinkaid1 first described the syndrome in three women with painful hips during the last trimester of their pregnancy. TO is a poorly recognized entity and the exact etiology of this condition remains unknown. It is characterised by progressive onset of pain in the joints of the lower extremity, accompanied by distinct findings of periarticular moderate to severe osteopenia. TO usually affects middle-aged men2, and women during the third trimester of pregnancy, or in the early postpartum period3. However, cases of non-pregnant women have also been reported4. TO mainly affects the hip, following knee, foot and ankle, while cases of bilateral localisation are rarely reported in the literature2,5.

Although, there are many published data of TO where both hips are affected, there are only very few reports of bilateral TO of the knees during pregnancy6-8. The aim of this study is to present a case of a pregnant woman with simultaneous TO in both knees, undiagnosed until parturition.

Case Report

A 40-year old woman was hospitalized in the obstetrics department of our hospital, in early postpartum period, with pain in both knees and inability of walking since the 6th month of her pregnancy. The patient reported progressive pain in left knee during the 25th week of pregnancy, followed by pain of the right knee one week later. The patient reported no history of injury or pathological findings of her knees in the past. Pain worsened and she remained bedridden during the last trimester of pregnancy.

Clinical evaluation revealed remarkable atrophy in both quadriceps, intense tenderness on palpation of femoral condyles and a restricted, painful range of motion from full extension to 20° of flexion. Blood test, including hematological and biochemical analyses, rheumatological profile and hormonal tests of thyroid and parathyroid were normal. C-Reactive Protein was elevated (16.29 mg/dl, normal values: 0-1.00), finding which was expected due to early post-surgical period (caesarean delivery).

Plain X-rays showed areas of decreased bone density in both patellas, femoral condyles and the anterior part of tibial.

Abstract

Transient osteoporosis is a rare self-limited condition that affects mostly middle-aged men or women in the third trimester of pregnancy. The most commonly affected joint is the hip, while bilateral involvement of the knees is extremely rare. Complete resolution of symptoms and MRI findings occurs over 8 to 10 months after clinical onset. We present a case of a 40-year-old woman diagnosed with bilateral transient osteoporosis of the knees, in the early postpartum period treated for 6 months with calcitonin, analgesics and physiotherapy.

Keywords: Transient osteoporosis, Knee, Pregnancy

The authors have no conflict of interest.
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condyles in both knees (Figure 1). Also, an MRI scan was performed in both knees (Philips Gyroscan T5-NT). Pulse sequences included sagittal and coronal, 4 mm thickness, T1 and T2 weighted spin echo images and proton density weight pulse sequences with and without fat-saturation. MRI revealed bone marrow oedema, in the lateral tibial condyles, medial and lateral femoral condyles and patella of the left knee. In the left knee, there was extensive bone oedema in the lateral femoral condyles, and patella as well (Figure 2). Moreover, in her left knee, a focal area of abnormal signal density was detected in the lateral femoral condyle, which it seemed like avascular necrosis (Figure 3). There was no cortical disruption in both knees.

Based on the above findings, the diagnosis of bilateral transient osteoporosis of pregnancy was established. Patient was treated with intermuscular injectable calcitonin, oral paracetamol and physiotherapy. A follow-up MRI was performed at 4 months after first presentation to the department (7 months since the onset of symptoms), in which improved signals in both knees, including the focal area of possible avascular necrosis were demonstrated (Figure 4). Patient showed further improvement in the 6-month follow-up, regaining normal range of motion and the ability to walk without pain.

Discussion

The aetiology of TO is still unknown. Although, several theories have been proposed for TO such as obturator nerve compression during pregnancy, demineralisation...
due to functional impairment, and viral infection, neither of them have been confirmed\textsuperscript{1,9}. A nontraumatic type of reflex sympathetic dystrophy has been also proposed as a theory\textsuperscript{10}, while other authors consider TO as an early reversible phase of avascular necrosis with nearly the same pathway pattern\textsuperscript{11}. Pregnancy may be the cause of such events mainly due to two reasons. First of all, pregnancy constitutes a stressing factor on maternal calcium homeostasis and also it appears to be associated with an increased risk of joint damage due to venous stasis\textsuperscript{4}.

Patients usually complain of progressive pain in affected joint. The range of joint motion is diminished. As already described, and confirmed in our study, symptoms of TO in the knee cause remarkable atrophy in adjacent muscles due to loss of mobilization\textsuperscript{7,8}. The physical history of the syndrome consists of three phases\textsuperscript{12}. The first phase

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**Figure 3.** Sagittal T2 (left) and sagittal T1 (right) weighted images of left knee, showing a focal area of abnormal signal density in lateral femoral condyle.

**Figure 4.** Coronal T2-weighted images of left (left) and right (right) knee during 5-months follow-up, showing improvement of signal density in affected area and effusion effect in both femoral and tibial condyles.
lasts approximately one month, and is characterized by progressive pain and functional impairment of the affected joint. The next two months comprise the second phase with unchanged symptoms and marked osteopenia demonstrated on X-rays. The last stage is the one of remodeling with improvement in bone density and may last up to 8 months. Our patient, was already in the second phase of TO when the diagnosis was established, confirmed by the radiographic pathological findings and by the fact that the onset of clinical symptoms was 3 months ago.

During clinical presentation of TO, radiographic bone density is affected after 6 to 8 weeks and periarticular diffuse osteopenia is apparent in X-rays. The subchondral bone is less affected and joint space remains normal\(^1^2\). Bone scanning may show an increase of radionuclide uptake in the affected joint just a few days after the symptoms start, but the low specificity of this exam should be taken into consideration\(^1^3\). Since both X-rays and bone scans are not allowed during pregnancy, MRI appears to be the gold standard for the diagnosis. It can detect changes as soon as bone scan and regression of abnormalities can be seen with improvement of symptoms\(^1^4\). In T1-weighted images present as decreased signal intensity and in T2- weighted images as increased signal intensity. MRI abnormalities and the presence of effusion can be attributed to bone marrow oedema, which is the characteristic pattern of TO\(^1^5\). MRI also constitutes the most useful tool to differentiate TO from Avascular Necrosis (AVN), which is described as a focal, well demarcated lesion in subchondral area and as a double-line sign in T2-weighted images. Although, there is no strong evidence to support relationship between TO and AVN\(^1^6\), in two available reports of TO of the knee\(^1^7\), including our study there was coexistence of MRI findings of bone marrow edema and an area of suspicious AVN in femoral condyles. Apart from AVN, other pathologies such as rheumatoid arthritis, osteoarthritis, crystal-induce arthritis and septic arthritis should be excluded\(^1^8\). TO must also be differentiated from metabolic bone diseases, primary bone tumor, metastases, bone infections (osteomyelitis, tuberculosis) and reflex sympathetic dystrophy\(^1^9\). The differential diagnosis from all of them can be made with usual laboratory exams and characteristic MRI findings.

The treatment of TO is conservative with symptomatic therapy, protected weight bearing and repeated MRI every three months\(^2^0\). Calcitonin, bisphosphonates, calcium, vitamin D supplements and physiotherapy can also be used\(^6,7\). Some authors recommend surgical treatment of TO of the hip with core decompression\(^2^0\), while others suggest surgical treatment only for the complicated cases with pathologic fractures. However, this technique has minimal therapeutic value for TO of the knee.

**Conclusion**

Until now, the exact mechanism of how pregnancy affects TO is still unclear. The rareness of this syndrome is reflected in the absence of studies with large numbers of cases. The bilateral localization of TO in knees constitutes an uncommon condition. MRI has the key role in diagnosis. Even though medical treatment can not affect the natural progression of TO, analgesics and functional therapy are essential to prevent fragility fractures.

**References**