



Idiopathic Transient Osteoporosis of the Hip in a Non-Pregnant Woman: A Case Report

Gebe Olmayan Bir Kadında Kalçanın İdiopatik Geçici Osteoporozu: Vaka Çalışması

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Summary

Transient osteoporosis of the hip (TOH) is a rare disease affecting women in the third trimester of pregnancy as well as middle-aged men. TOH is associated with hip pain and temporary osteopenia apparent on radiology without joint space narrowing or destruction of the hip. Pregnancy is the only recognized risk factor for women. It typically runs a benign course with eventual resolution of symptoms. Radiographs may be unrevealing early in its course. Therefore, transient osteoporosis may be confused with many other conditions such as avascular necrosis. In early stages, it is important to distinguish between these two conditions to order to decide on an appropriate treatment plan. In this report, we present a 35-year-old woman who had TOH, without any risk factor including pregnancy, and showed sufficient improvement with conservative treatment. *Türk J Phys Med Rehab 2013;59:157-60.*

Key Words: Transient osteoporosis, hip, pregnancy

Özet

Kalçanın geçici osteoporozu (KGO) öncelikle orta yaşlı erkekleri ve gebeliğin üçüncü trimesterindeki dönemindeki kadınları etkileyen nadir bir hastalıktır. KGO kalça ağrısı ve radyolojik olarak eklem aralığında daralma veya destrüksiyon olmaksızın ortaya çıkan geçici osteopeni ile ilişkilidir. Kadınlar için tanımlanmış tek risk faktörü gebeliktir. Tipik olarak semptomların kendiliğinden rezolüsyonu ile benign bir seyir gösterir. Radyografiler erken dönemde belirti vermeyebilir. Bu nedenle, geçici osteoporoz avasküler nekroz gibi çok sayıda diğer hastalıklarla karışabilir. Erken dönemlerde bu iki hastalığın birbirinden ayırtılması uygun tedavi planının seçilebilmesi için önemlidir. Bu yazıda, gebelik de dahil olmak üzere herhangi bir risk faktörü olmayan ve konservatif tedavi ile yeterli iyileşme gösteren KGO'lu 35 yaşındaki kadın hasta sunulmuştur. *Türk Fiz Tıp Rehab Derg 2013;59:157-60.*

Anahtar Kelimeler: Geçici osteoporoz, kalça, gebelik

Introduction

Transient osteoporosis of the hip (TOH) is a condition of unknown etiology, and is often self-limiting. Characteristic features of the disease are disabling pain in one or more joints and radiographical evidence of osteopenia limited to involved joints. TOH is a rare cause of hip pain. The syndrome is relatively infrequent and radiographic findings can not be determined in early stages (1,2). Radiographic examinations performed approximately one month after the onset of the symptoms demonstrate demineralization of the femoral head and neck without involvement of the joint space, For this reason, transient osteoporosis may be confused with many

other conditions such as avascular necrosis. In the early stages, the distinction between these two conditions is important in order to decide on an appropriate treatment plan (2-4).

Transient osteoporosis of the hip usually develops in middle-aged men and in women in the third trimester of pregnancy. Typically, patients are initially seen with acute, progressive pain in the hip, an antalgic gait, and a functional disability involving the affected extremity (5). The disease is self-limiting, however, within a few months (6 to 8 months), the pain as well as the radiological abnormalities disappear spontaneously, with complete resolution of the condition (6,7).

In this report, we present a rare case of TOH in a non-pregnant middle-aged woman.

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Case Report

A 35-year-old woman was admitted to our outpatient clinic with pain on the right hip for 10 days. She did not have any systemic disease and was not taking any medication. There was no history of trauma, morning stiffness, weight loss and fever. She had no history of smoking and alcohol use. Family history was negative for osteoporosis or its symptoms. She had three sons, and had given birth to the last one five years ago. She was not pregnant in the meantime.

The pain was localized to the right groin and was typically aggravated by weight bearing and relieved by rest. There was no pain in any other joint. Musculoskeletal examination revealed minimal groin tenderness of the right hip. Abduction and external rotation of the right hip was limited due to pain. Her gait was antalgic. Other range of motion (ROM) values of all joints were within normal limits; muscle strength and neurological examination were also normal.

Laboratory tests revealed complete blood count, erythrocyte sedimentation rate, rheumatoid factor, C-reactive protein, Brucella Ig test, thyroid, kidney and liver function tests, serum calcium, phosphorus, magnesium, alkaline phosphatase, 25(OH)D³ and parathyroid hormone within normal limits. Radiograph of the pelvis revealed normal findings (Figure 1). Magnetic resonance imaging (MRI) showed low signal intensity in the femoral head and neck on T1-weighted images (Figure 2a) and homogeneous high signal intensity on T2-weighted images, with joint effusion and joint space preservation (Figure 2b). However, the patient refused scintigraphic examination, the available imaging findings, clinical and laboratory examinations were consistent with the diagnosis of TOH.

She was treated with nonsteroidal anti-inflammatory drug (NSAID) (indomethacin, 100 mg/day for one month) and ROM exercises for two weeks, non-weight bearing crutch walking and abductor muscle strengthening exercises were also ordered. Three months later, the patient showed more than 50% improvement in both symptoms and ROM of the right hip. This benign clinical course also supported the diagnosis of TOH in our patient.

Discussion

Transient osteoporosis of the hip is an uncommon, usually self-limited, distinct clinical entity of unknown etiology. Curtiss and Kincaid described the first 3 cases in 1959, all of which involved pregnant women in their third trimester who presented with thigh pain exacerbated by weight bearing with radiographic evidence of femoral head osteopenia in the absence of any systemic metabolic disturbance (8). The condition was named transient osteoporosis of the hip by Hunder and Kelly in 1967 (9).

Two thirds of the cases are reported in healthy middle-aged men between the ages of 40 to 60 years. One third of the cases occur frequently in women who are in the third trimester of pregnancy or in the early postpartum period. Children are rarely affected (1,3). Although the hip is most commonly affected; transient osteoporosis can affect the knee, foot, ankle and less frequently shoulder, lumbar spine, elbow and the wrist. The left hip is involved more commonly than the right,

with bilateral involvement occurring in 25-30% of patients (10). The only recognized risk factor for bilateral involvement is pregnancy (1,2,10). In our case, the right hip was involved and she was not pregnant.



Figure 1. Radiograph of the pelvis (normal radiologic findings). Intensity in the femoral head and neck on T1-weighted images.

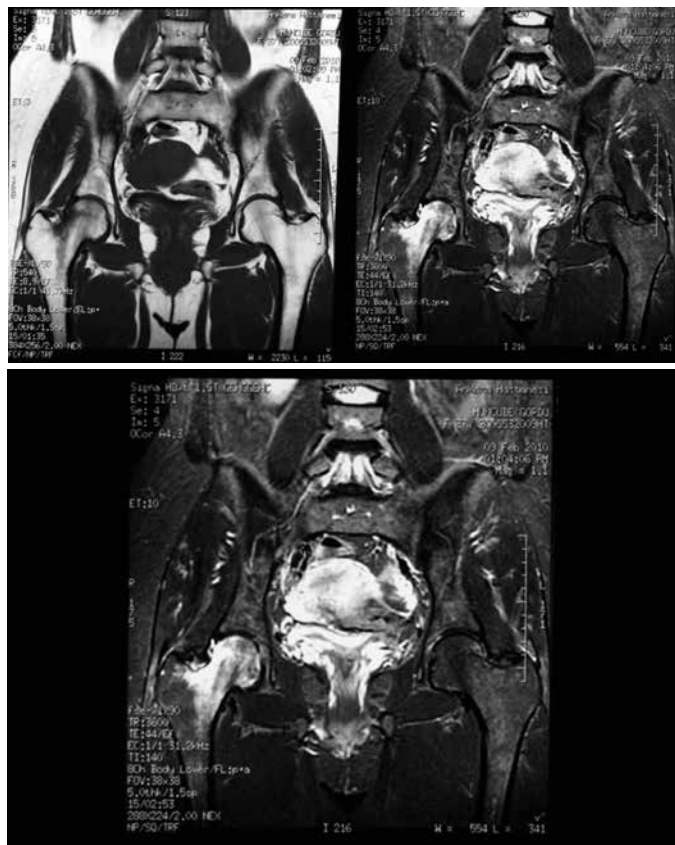


Figure 2. Magnetic resonance imaging of the hip-Low signal intensity in the femoral head and neck on T1-weighted images (Figure 2a) and homogeneous high signal intensity on T2-weighted images, with joint effusion and joint space preservation (Figure 2b).

Dull ache in the inguinal area, buttocks, or anterior aspect of the thigh is the presenting symptom of transient osteoporosis, and is usually acute in onset. It is frequently accompanied by an antalgic gait. Patients often remember the time of the beginning of the symptoms. The pain is exacerbated by weight-bearing and relieved by rest. Night pain is rarely seen. Physical examination of the hip may be slightly tender or totally normal. ROM is generally preserved, but occasionally there may be a slight decrease of abduction and rotation due to hip pain (2,11). In our case the presenting symptom was acute onset right hip pain. Physical examination revealed mild groin tenderness and minimal reduction of abduction and external rotation.

The precipitating factors and pathogenesis of TOH remain elusive. Implicating factors include genetic predisposition, compression of the obturator nerve, Sudeck's atrophy, bone medullary hypertension and small vessel ischemia, and chemical or hormonal factors related to pregnancy (10). The association with the third trimester of pregnancy is intriguing. Several factors seem to predispose women to TOH during pregnancy, such as negative calcium balance, increased activity of the adrenal cortex and an increased demand for protein and minerals. But these hypotheses, however, does not fully explain the selective demineralization seen in this entity. In addition, venous retention due to impaired venous return and leading to medullary hypertension has been proposed as a contributing factor for the development of TOH in pregnancy (1,12).

The differential diagnosis must include avascular necrosis, septic arthritis and malignancies. Additionally, inflammatory joint diseases, fractures of the femoral neck, reflex sympathetic dystrophy, pigmented villonodular synovitis and synovial chondromatosis should also be taken into consideration (9,13,14). History, physical examination, radiological analysis, laboratory tests and clinic course of the condition are also important in diagnosis. In our patient, laboratory tests were within normal limits and there were not any systemic finding. On MRI, the extension of the signal intensity changes from the metaphysis to the epiphysis also favors TOH as in our case, and differentiates the disease from malignancies and septic arthritis which are usually located in the metaphysis (2,6,14).

The main challenge lies in distinguishing between TOH and AVN. Some authors suggested that transient ischemic insult to the bone may be cause of transient osteoporosis. In this theory, it is postulated that the ischemic insult results in only limited cell death involving only the hematopoietic and fatty elements. However, in AVN, the cell death includes osteocytes. As a result, there may be a spectrum of ischemia, from a more limited insult resulting in transient osteoporosis to a more extensive affront progressing to bone death and AVN. According to this, TOH corresponds to early changes of AVN, but such an accelerated repair inhibits the progression to AVN (2,15,16).

The differential diagnosis between TOH and AVN can be difficult, especially at an early stage. This is important because the prognosis of AVN can be unfavorable. There are several distinguishing clinical and radiographic features that distinguish between these two clinical entities. TOH is classically characterised by an acute onset disabling pain, which is exacerbated by weight bearing and relieved by rest. Antalgic

gait is usually present. Range of motion is usually preserved with occasional restrictions on rotation and abduction. TOH is generally a self-limited condition that responds to treatment whereas AVN is usually progressive. In AVN, the pain is typically exacerbated by weight-bearing, but it is often present at rest. The limp and an antalgic gait are typically late findings. Usually, the pain becomes more severe and physical examination reveals restriction of movement, especially after the collapse of the femoral head. Patient may have any of the risk factors (such as steroid intake, alcoholism, trauma and haemoglobinopathies) (2,11). None of these risk factors were present in our case and also she was not complaining of pain at rest.

Plain radiographs show changes 3 to 8 weeks after the onset of symptoms in both TOH and AVN. The involvement in TOH usually diffuses into the proximal femur (head-neck), and the lesion is homogeneous. The trochanters, acetabula, iliac wings and ischiopubic rami may be affected. The joint space is always preserved and osseous erosion or subchondral collapse is never observed. In AVN, there is no homogeneity, and the involvement is segmental or focal in the anterosuperior (subchondral) region of the femoral head. Additionally, the subchondral collapse (sign of half-moon), pathognomonic for avascular necrosis which particularly develops in the advanced phases, is not seen in TOH (3,6,14). Since our patient was in the early stage of the disease, plain radiography of the pelvis revealed normal findings.

MRI shows abnormal findings before conventional radiography, as early as 48 hours after the onset of TOH (14). Edema and fat necrosis in the femoral region result in a reduced T1 signal, whereas T2 signals are increased due to osteopenia extending from the femoral head to the intertrochanteric region which is diffuse and homogeneous as in our case. Early stage of AVN with bone marrow edema can mimic TOH features. It is difficult to differentiate from each other. However, it has been demonstrated that the initial MRI findings in AVN is a band pattern. Moreover, the absence of focal defects and subchondral changes on MRI is highly suggestive of TOH (14,17,18).

In TOH, the bone scintigraphy demonstrates a diffuse, homogeneous, increased uptake in the head and neck of the femur extending to the intertrochanteric line (1). In AVN, the bone scintigraphy may reveal similar findings, but increased uptake is limited to the femoral head and may be less intense. The scintigraphic feature of AVN is cold spot in hot area. Uptake of the isotope is decreased over the the anterosuperior region of the femoral head surrounded by an area of increased uptake due to reactive hyperemia. Cold spot in hot area is almost pathognomonic to AVN and is never seen in TOH (2,14,19). Scintigraphic examination was not applied in our patient. Radiographs, bone scans, MRI, characteristic symptoms and the benign self-limiting course will clarify the diagnosis of TOH. In our patient, the absence of common risk factors for AVN, acute onset, typical MRI findings and no clinical progression supported the TOH diagnosis.

Various treatments have been attempted in TOH. The aims of therapy are prompt relief of pain, acceleration of functional recovery of the affected joint and reduction of microfractures and prevention of pathological stress fractures. The treatment

consists of prolonged rest, protected weight bearing and the prescription of NSAIDs. ROM exercises should be started as soon as the patient is comfortable, in order to prevent contracture. Prolonged bed rest leads to abductor muscle weakness in these patients and thus, abductor muscle strengthening exercises are an important component of conservative treatment. In addition, oral, intravenous and intramuscular bisphosphonates have been reported to have beneficial effects (20,21). Calcitonin and prednisolone, have also been used. Calcitonin has been proposed to reduce bone resorption and to accelerate recovery (1,14,22). However, there are no well-controlled safety studies of bisphosphonates or calcitonin in TOH. In AVN, generally invasive operative intervention may be warranted (23). Our patient was treated with NSAIDs. ROM and abductor muscle strengthening exercises were also applied, eventually dramatic clinical improvement was achieved parallel to benign course of the disease.

In conclusion, TOH is a rare disorder with a typical presentation and a benign course. The pathogenesis is still unknown. The only recognized risk factor is pregnancy for women. However, as in our case, with acute onset hip pain, TOH should be born in mind even, if the patient was not pregnant, to avoid misdiagnosis especially from AVN. Early diagnosis will prevent unnecessary diagnostic procedures and therapies. Patients correctly diagnosed as having TOH usually represent sufficient improvement on conservative treatment.

Conflict of Interest

Authors reported no conflicts of interest.

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