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Case Report

**Transient Osteoporosis of the Hip during Pregnancy: A Case Report**

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Transient osteoporosis, also called “bone marrow edema syndrome”, is a rare disorder of unknown etiology. It primarily affects women in the third trimester of pregnancy as well as middle-aged, overweight men, and the hip joint is most commonly involved. The main symptom of transient osteoporosis of the hip is a sudden onset of pain at the lateral aspect of the hip. Mild limitation of hip range of motion is often present. A 36-year-old woman presented to our outpatient department 5 weeks after cesarean section with bilateral hip pain, which had begun suddenly during her eighth month of pregnancy. Magnetic resonance imaging of the pelvis revealed marked bone marrow edema at both femoral heads and necks. According to history, physical examination and image findings, transient osteoporosis of the hip was diagnosed.

Bone densitometry revealed osteopenia. The patient was begun on a regimen of analgesic medication and physical therapy, and she resumed ambulation 3 months after the cesarean section. Arriving at an accurate diagnosis of this disorder requires ruling out other possible causes of hip pain, including avascular necrosis, stress fracture of the femoral neck, and neoplasia. Early diagnosis is important to prevent possible complications such as fracture. (Tw J Phys Med Rehabil 2009; 37(2): 131 - 137)

**Key Words:** transient osteoporosis, bone marrow edema syndrome, magnetic resonance imaging, hip, pregnancy

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**INTRODUCTION**

Osteoporosis is generally a progressive and painless condition. In contrast, transient osteoporosis is a rare, self-limiting skeletal disease that is both reversible and painful. The most common site affected is the hip joint, with the left hip more frequently involved than the right. Bilateral involvement occurs in only 25-30% of cases. Up to 40% of patients have involvement of other joints, such as lower-extremity joints, upper-extremity joints, and the spine, with migration to multiple sites or recurrence seen in 25-50% of this population. The disorder primarily affects women in the third trimester of pregnancy, as well as middle-aged (40-50 years old), overweight men. In most cases, the first symptom is the sudden onset of pain. In transient osteoporosis of the hip, the pain is usually located at the lateral aspect of the hip, the front of the thigh, the buttocks, or the groin. There is usually no history of trauma to the joint that would trigger pain. Mild limitation of range of motion (ROM) of the hip is frequently noted, and the pain may
increases with turning movements or weight bearing and lessens with rest. The pain gradually increases over a period of weeks or months and may eventually become disabling. Alteration of gait may be observed as the patient tries to protect the joint and ease the pain.

Diagnosis is made by excluding other possible causes of hip pain. The clinical course is benign, and spontaneous recovery usually occurs over 2-12 months. In cases arising during pregnancy, the symptoms tend to resolve during the first few postpartum months.

In this article, we report a rare case of bilateral transient osteoporosis of the hip (TOH) during pregnancy.

**CASE REPORT**

A 36-year-old woman with no systemic disease experienced a sudden onset of bilateral hip pain during her eighth month of pregnancy. The pain caused the patient to alter her gait, but she was able to walk with minimal assistance. The visual analog scale (VAS) pain score at that time was 3-4 on a 10-point scale with 1 indicating the least pain and 10 indicating the worst pain. Bilateral limitation of hip ROM due to pain was also noted. This was the patient’s first pregnancy; she was carrying twins and had experienced a 15-kg increase in body weight during the pregnancy. The leg discomfort caused her to become virtually bedridden during her last month of pregnancy. The pregnancy was otherwise uncomplicated.

The patient underwent cesarean section at another hospital because of fetal positioning. Subsequently, the hip pain worsened, with the VAS pain score increasing to 7-8. Initially, an orthopedist of that hospital was consulted, and bilateral hip radiography was performed, which revealed no specific findings. The patient was then referred to the physical medicine and rehabilitation outpatient department of that hospital, where she underwent physical therapy modalities including interferential current therapy, shortwave diathermy, and transcutaneous electrical nerve stimulation. Her VAS pain score was around 3-4 after treatment. Although her hip pain improved, she still could not go up and down stairs after returning home.

The patient came to our outpatient department 5 weeks after the cesarean section, with persistent bilateral hip pain that was worse on the right side. Neurological examination revealed bilateral lower-extremity muscle weakness (hip flexor: 3/3; knee extensor: 4-/4; ankle extensor: 5/5), normal deep tendon reflexes, and intact sensation. Physical examination showed positive Patrick’s test bilaterally and no limitation of hip ROM. The patient used a wheelchair for all of her activities. Her functional independence measure (FIM) was 85 at that time. She need moderate assist in transfer and ambulation. There was no history of trauma, fever, or body weight loss. Basic laboratory data were checked to confirm or rule out myopathy or avascular necrosis; pelvic magnetic resonance imaging (MRI) and nerve conduction velocity (NCV)/electromyographic (EMG) examination were also arranged.

Laboratory data and NCV/EMG findings were normal. The erythrocyte sedimentation rate (ESR) was 61 mm/hr; the C-reactive protein (CRP) level was 1.93 mg/dL. MRI of the pelvis revealed marked bone marrow edema at the femoral head and neck bilaterally and mild hip joint effusion bilaterally (Figure 1. A-C). According to history, physical examination and image findings, transient osteoporosis of the hip was diagnosed.

The patient was begun on a regimen of analgesic medication and physical therapy, including strengthening exercises and postural training. Her hip pain improved, as did her functional status from sitting to standing. Bone densitometry was performed 11 weeks after the cesarean section and revealed osteopenia (T-score of lumbar spine: -1.24; left hip: -2.16; right hip: -2.48). The results of bone densitometry confirmed the diagnosis, but it was not the main diagnostic tool, and the results were above the range of osteoporosis because it was taken 11 weeks after the cesarean section. She resumed ambulation and independent daily activities 3 months after the cesarean section. Her FIM score improved to 118 at that time. Follow-up bone densitometry performed 5 months after the cesarean section showed slight improvement (T-score of left hip: -1.83; right hip: -1.76).

**DISCUSSION**

Transient osteoporosis was first documented in 1959
by Curtiss and Kincaid, who called it “transient demineralization”. The term “transient osteoporosis” was first used in 1968 by Lequesne. The condition is also called “bone marrow edema syndrome” because of the MRI findings early in the progression of the disorder. Other terms that have been used include “regional migratory osteoporosis,” “transient migratory osteoporosis,” and “hip algodystrophy”.

The etiology of this disorder is still unknown. Among the pathogenetic factors that have been proposed are neural, vascular, chemical, and hormonal mechanisms. Possible neural mechanisms include a variant of reflex sympathetic dystrophy, pathology of the peripheral nerves, compression of the obturator nerve, and irritation or compression of the pelvic nerves. Suggested vascular mechanisms include ischemia of the small vessels proximal to the nerve roots, obstruction of venous return, transient ischemia, and decreased fibrinolytic potential.

Proposed chemical and hormonal mechanisms include factors associated with pregnancy and increased maternal demands for calcium, protein, and minerals. Transient osteoporosis may also be associated with liver cirrhosis, disorders related to increased bone turnover, or hypercalciuric osteoporosis. Genetic predisposition and

Figure 1. (A) Magnetic resonance images (MRI) of pelvis showing marked bone marrow edema at femoral head and neck bilaterally (arrows), mild bilateral hip joint effusion, no definite abnormal signal at soft tissues bilaterally, and no definite osteolytic lesion. (B) MRI of pelvis (T1-weighted image) showing decreased signal intensity of bone marrow at femoral head and neck bilaterally (arrows). (C) MRI of pelvis (T2-weighted image) showing increased signal intensity of bone marrow at femoral head and neck bilaterally. (arrows)
Transverse osteoporosis occurs in three phases: (1) Stage 1 (first 1-2 months) features a rapid increase in pain and functional disability but normal radiographic findings; (2) Stage 2 (after 2-3 months) features maximal symptoms and signs, with osteopenia noted on radiographs; (3) Stage 3 (up to 6 months) involves gradual regression of the disease and radiographic changes, the latter taking longer.[4,14] Magnetic resonance imaging also shows three stages-diffuse, focal, and residual findings-consistent with the three clinical stages.[13] The present case suffered from bilateral hip pain since her eighth month of pregnancy. Maximal pain was noted after cesarean section, but radiographs did not reveal osteopenia at that time. Her pain gradually improved in the following 2 months. MRI revealed bone marrow edema and bone mineral density (BMD) revealed osteopenia. Follow up BMD 5 months after cesarean section shows improvement in bone marrow density.

Diagnosis is made by exclusion of other possible causes of hip pain.[2] The most important differential diagnosis is avascular necrosis, in which the pain has an insidious onset and typically occurs at rest.[3] Transient osteoporosis is not an early reversible form of avascular necrosis.[5] Other differential diagnoses include reflex sympathetic dystrophy, stress fracture of the femoral neck, inflammatory arthritis, septic arthritis, synovial disorders (pigenated villonodular synovitis, synovial chondromatosis), and neoplasia (carcinoma, multiple myeloma).[11,12] Unlike transient osteoporosis, reflex sympathetic dystrophy typically features pain at rest, is associated with cutaneous vasomotor changes, and can have articular and osseous sequelae.[3,12,15,16]

Physical examination of a patient with transient osteoporosis of the hip reveals only limitation in ROM and a positive Patrick’s test.[6] Laboratory data are typically within the normal ranges, except for an elevated ESR, but this is a common finding during pregnancy.[8] Imaging can help in the diagnosis of this condition. Plain radiographs of the hip may not show radiolucency until the condition is advanced (usually 1-2 months after the pain starts).[2,7] Remineralization usually occurs within 2 years.[7] Transient osteoporosis may be confused with avascular necrosis or a femoral neck stress fracture, but the joint space is preserved.[2,3,6] A bone scan is sensitive but not specific.[7] Increased diffuse uptake of the whole femoral head, neck, and acetabulum is noted in the first few days after the onset of symptoms, even before the radiographic changes are seen, and returns to normal 12-15 months after the onset of symptoms.[7] Computed tomography should be used only as a complementary investigative tool.[7] It shows the joint space integrity and the extent of local demineralization and excludes other pathological conditions.[7]

MRI is the best tool currently available for detection of transient osteoporosis.[2,5,7] It typically shows bone marrow edema within 48 hours after the onset of symptoms and resolution within 6-8 months.[1,5,7] It shows decreased signal intensity of bone marrow on T1-weighted images, increased signal intensity of bone marrow on T2-weighted images, and surrounding soft-tissue edema and joint effusion on T2-weighted images.[1,5] Differential diagnosis of MRI bone marrow edema includes ischemia, avascular necrosis, reflex sympathetic dystrophy, inflammation, osteomyelitis, rheumatoid arthritis, trauma, bone bruise, stress fracture, neoplasm, and other causes.[5,7] If the patient is pregnant, the image studies should be delayed until the last stages or after delivery.[8] MRI of the present case revealed bone marrow edema. Normal range of laboratory data and negative finding in plain film excluded the possibility of other differential diagnoses. Bone densitometry shows 3-24% bone loss after 3-5 months from the onset of symptoms, with a return to normal bone density within 2 years.[4,9] BMD of the present case shows improvement in 5 months. This improvement proved our diagnosis, but it was not the main diagnostic tool.

The clinical course is benign, with spontaneous recovery usually occurring over 2-12 months for a male or 2-6 months for a female after pregnancy.[1] Prevention of damage to bones weakened by osteoporosis is important; the most severe possible complication is stress fracture of the hip.[1,9] Fracture risk can be predicted according to three factors: (1) the rate of bone loss of the femoral neck, (2) the baseline BMD at the femoral neck, and (3) ad-
vancing age. The highest fracture risk has been reported to be at a mean of 8.3 weeks, when the lowest bone mineral density was noted.\[9\]

Treatment is mainly conservative.\[3\] Symptomatic treatments include reduction of weight bearing activities, analgesic medication, and physical therapy.\[3\] Use of crutches, a cane, or other walking aids helps relieve the stress of weight bearing. Nonsteroidal anti-inflammatory drugs can be administered to relieve pain. Physical therapy, including exercises to increase flexibility and ROM, is helpful; the use of traction has been reported for a male patient.\[4\] Prolonged bed rest leads to profound abductor muscle weakness in these patients and thus, abductor muscle strengthening exercises are an important component of conservative treatment.\[17\] The rehabilitative goals of the present case were reducing bilateral hip pain, preventing stress fracture, preventing hip joint contracture, improving muscle strength of bilateral low extremities, and improving functional status. Rehabilitation program including ROM exercise, postural training, strengthening training and ambulation training was given.

Pharmacological treatments such as bisphosphonates and calcitonin can also help relieve pain.\[4,12,18\] Surgical treatment such as core decompression is controversy for this benign process.\[3,12,18\] No treatment has been shown to have any effect on the actual disease process.\[1,7\]

Elective cesarean section and cessation of breast-feeding may relieve the symptoms, but there are no controlled studies to support these suggestions.\[2,4,6,19\] The present case did not improve after cesarean section. On the other hand, her pain increased after cesarean section, which is not the documented course in the literature. Modalities had been given before her visit to our OPD which showing some improvement in pain, but her functional status improved little. We gave NSAID and physical therapies including therapeutic and strengthening exercises and postural training. These indeed show improvement in both her pain and functional status.

Aches and pains in various parts of the body are common complaints of the pregnant woman, and complaints of hip pain are relatively frequent.\[10\] Transient osteoporosis of the hip should be considered in the differential diagnosis of hip pain in pregnant women, as well as in middle-aged, overweight men. Unlike osteoporosis, transient osteoporosis is a painful and reversible condition. Both disorders require early diagnosis to prevent complications such as hip fracture.\[1\] Transient osteoporosis is self-limiting, with spontaneous resolution within 1 year in the majority of cases.\[3\] Consequently, invasive investigations are rarely necessary.\[3,5\]

There have been more than 300 cases of this disease reported in the literatures.\[9\] However, the disease appears to be rare in Asians, and few cases have been reported from Asian countries.\[17\] To our knowledge, this is the first case reported in Taiwan.

**CONCLUSION**

Transient osteoporosis of the hip should be considered in the differential diagnosis of hip pain in pregnant women, as well as in middle-aged, overweight men. Early recognition and appropriate management are important to prevent damage to weakened bones and help relieve patients’ fear of lifelong disability.

**REFERENCES**


懷孕期間髖關節一過性骨質疏鬆症：病例報告

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懷孕期間的髖關節疼痛並非罕見的問題，但疼痛程度足以影響行走的卻是少數。本文報告一位36歲的健康婦女，她在孕期的第8個月開始感到兩側的髖關節疼痛，並影響到步行的能力。產後疼痛的情況加重，照過X光之後確定沒有骨折等問題，抽血檢查以及肌電圖檢查都得到正常的結果，在懷疑缺血性壞死(vascular necrosis)的情況下安排了髖關節的核磁共振造影(MRI)檢查，結果發現兩側股骨頭及股骨頸的骨髓水腫(bone marrow edema)，符合一過性骨質疏鬆症(transient osteoporosis)的診斷。這個症狀首先在1959年被提出。其致病原因不明。主要出現在兩個主要的族群，其一是懷孕最後三個月的婦女，其二是過重的中年男性。症狀通常開始於髖部的疼痛，之前並無外傷，體檢可能會發現輕微的關節活動限制。其診斷的確立要先排除其他可能的髖部疼痛原因，例如缺血性壞死，骨折以及腫瘤。核磁共振造影是最好的診斷工具。這是一種自限性的疾病，通常會在2到12個月之內自行好轉。目前尚無任何藥物或治療確可以影響其病程。骨折是其最嚴重的併發症，故減輕患側的負重，並避免在骨質減少的期間產生骨折是應該注意的地方。這種罕見疾病在亞洲地區尤其少見，這是台灣首位被報告的病例。（台灣復健醫誌 2009；37(2):131 - 137）

關鍵詞：一過性骨質疏鬆症(transient osteoporosis)，骨髓水腫症候群(bone marrow edema syndrome)，核磁共振造影(magnetic resonance imaging)，髖關節(hip)，懷孕(pregnancy)