Letters to the Editor

Transient Osteoporosis of Both Hips in Pregnancy

Osteoporosis transitoria de ambas caderas en el embarazo

To the Editor,

Transient osteoporosis of the hip (TOH) can occur in the context of pregnancy. In this case, it often develops in the last trimester and, to a lesser extent, before or after labor. It is usually unilateral and most likely to occur on the left side.\(^1\)\(^{-3}\) Because it is uncommon, we present the case of bilateral TOH, with greater involvement of the right side, during the fifth month of pregnancy.

The patient was a 34-year-old woman undergoing her first pregnancy with a history of idiopathic left hip TO, which occurred six years earlier. During the fifth month of pregnancy and with no other known trigger, she began to have pain in the right buttock which was attributed to a pyramidal bone syndrome. Afterwards, the pain was located on the outside of the right hip and was classified as a probable trochanteritis. In the absence of improvement, she was referred to rheumatology during the seventh month of pregnancy. She complained of pain on the lateral side of both hips, more markedly on the right, accompanied by significant functional impairment. This pain was compounded by weight burden and gait, forcing her to use crutches. The examination revealed pain and limitation in abduction and external rotation of the right hip, without alterations in other areas of the musculoskeletal system. Otherwise, laboratory data did not see an increase in acute phase reactants, with negative rheumatoid factor, citrullinated peptide antibodies and HLAB27. There was no radiological or scintigraphic study performed due to the pregnancy. Suspecting a TOH, she was treated with paracetamol, physiotherapy and unloading of the joint. During the postpartum period, we performed an X-ray which showed no abnormalities, and an MRI in which “intraspynocephalic and intertrochanteric right and left marrow edema, with no signs of osteonecrosis or sacroiliitis, all of this indicating a bilateral TOH” (Fig. 1). After the birth, she had progressive improvement and the symptoms disappeared after 2 months. In a second imaging control, at eighth months postpartum, there was a restoration of normalcy in the MR images (Fig. 2).

Prevalence of TOH due to pregnancy is difficult to assess. It usually occurs in the last third of pregnancy and to a lesser extent, before or postpartum. Its pathogenesis is unclear and, therefore, involves several factors: microtrauma, trabecular microfractures by decreased bone mass during pregnancy, circulatory changes characteristic of pregnancy and compression of the obturator or pelvic sympathetic nerve by the gravid uterus. Clinically, it usually presents with groin pain or pain adjacent to the hip, of a mechanical rhythm, accompanied by functional impairment. The

Fig. 1. MRI Indicative of a Bilateral TOH.

Fig. 2. Restoration of Normality on MRI.
preference for the left hip is allegedly due to cephalic presentation of the fetus, which could lead to repeated microtrauma in that area. Bilateral involvement is exceptional. Similarly, migratory forms have been described affecting the knee, ankle or foot. Laboratory changes only relate to those seen during pregnancy. Initially, X-rays are normal and later (1–2 months), a homogeneous demineralization, that does not affect the joint line may be seen. Early on scintigraphy shows increased uptake of isotope in the acetabulum and femur. MRI is essential for diagnosis, showing edema in the affected area and ruling out osteonecrosis, as in the present case. It is convenient to make a differential diagnosis with other hip problems (osteonecrosis, inflammatory rheumatic disease, infectious disease, metabolic disease, synovial disease, neoplasia, osteomalacia and trauma) or surrounding area disorders (lumbar, sacroiliac, symphysis pubis, uro-genital and digestive). Treatment is based on the joint unloading, analgesia and physical therapy. It usually has a favorable outcome and recovery without sequelae in variable periods of time (2 months to 1 year). However, some cases leading to osteonecrosis or femoral neck fracture have been described.1–8

In sum, we consider it necessary to rule out the presence of primary TOH when there is groin or hip pain in a pregnant woman. Similarly, early diagnosis is important using scintigraphy and MRI when circumstances permit. In addition, early treatment is essential for a quick recovery. Finally, it is important to monitor a possible progression toward osteonecrosis or fracture of the femoral neck.

References

Carlos Bruscas Izú. a, b * Sara San Juan de la Parra b

a Servicio de Reumatología, Hospital de la Defensa, Zaragoza, Spain
b Servicio de Alergología, Hospital de la Defensa, Zaragoza, Spain

* Corresponding author.
E-mail address: carlosbruscas@hotmail.com (C. Bruscas Izú).

Acute Meningitis in Behcet’s Disease

Meningitis aguda en la enfermedad de Behcet

Dear Editor,

Behcet’s disease (BD) is a chronic relapsing occlusive vasculitis of unknown etiology, characterized by the presence of oral and genital ulcers and intraocular inflammation, with less common cutaneous, joint, vascular, digestive and neurological involvement.1 The presence of neurological symptoms in the context of BD constitutes what is called neuroBehcet’s, characterized in most cases by the presence of aseptic meningitis, cerebellar signs, intracranial hypertension, and pyramidal alterations.2 We report the case of a 34 year old male with a history of BD, who presented an altered consciousness and meningeal signs, diagnosed with neuroBehcet and showing a favorable clinical response to treatment with triple immunomodulatory therapy.

The patient is a Spanish man of 34 years with BD, in whom the diagnosis was established by the presence of oral and genital ulcers and repeated episodes of anterior uveitis three years prior, treated with prednisone at a dose of 5 mg/day and cyclosporin A. He came to the emergency department 24 h before due to a self-limited episode of loss of consciousness and subsequent headache and vomiting. The general physical examination and vital signs were normal except for the presence of nuchal rigidity upon neurological examination. There were no signs of disease activity at the time of the initial evaluation.

He was put in reverse isolation for suspected intracranial meningeal syndrome secondary to infection due to immunosuppression. During observation in the emergency department a blood count was performed which showed 4130 WBC E9/l and no left shift, hemoglobin 11.5 mg/dl, hematocrit 36%, ESR 12 mm/h, lactate dehydrogenase 97 U/L, total protein 6.2 g/l albumin 3 g/l, and a computed tomography that revealed no significant structural alterations.

In accordance with the headache and meningeal signs a lumbar puncture was performed, resulting inconclusive due to a traumatic technique, and no second sample was obtained. However, the patient was admitted to the neurology department with empirical intravenous antibiotic therapy based on ceftriaxone and vancomycin as well as acyclovir for antiherpetic coverage.

During admission, he underwent a second lumbar puncture with glucose 56 mg/dl, protein 68 mg/dl and WBC 600/µl (90% polymorphonuclear cells, 10% lymphocytes). Due to the polymorphonuclear pleocytosis we performed a magnetic resonance imaging (MRI) scan (Fig. 1), which showed an extrusion affection with exten- sion to the left cerebral peduncle which appeared isointense on T1, hyperintense on T2 (Fig. 2), and flair, and hypointense on T1 IR.

Microbiological studies with Gram and Ziehl–Neelsen stains of the cerebrospinal fluid cultures (including Lownenstein), polymerase chain reaction for herpes virus, Brucella and Borrelia serologies were negative. We also excluded other possible differential diagnoses considering the history of immunosuppression: varicella zoster virus, herpes virus, pox, cytomegalovirus, Candida and meningeal lymphomatosis.

He was assessed by neurology who considered neuroBehcet given the persistence of symptoms despite antibiotic treatment and the medical history of the patient and who initiated infliximab therapy associated with tuberculosis prophylaxis with isoniazid, prior suspension of antibiotic treatment and cyclosporin A, the latter due to the relationship described in the literature with worsening neurologic manifestations.3,4

Regarding the imaging differential diagnosis it should be noted that, because of its topography and signal intensity, similar images