

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

- Curtiss PH, Kincaid WE. Transitory demineralization of the hip in pregnancy. A report of three cases. *J Bone Joint Surg.* 1959;41:1327–33.
- Lequesne M. Transient osteoporosis of the hip: a non traumatic variety of Sudeck's atrophy. *Ann Rheum Dis.* 1968;27:463–71.
- Bruscas Izu C, Calatayud Pérez JB, Gracia Galve A, Arregui Calvo R. Distrofia simpático refleja en el post-parto. *Ann Med Interna (Madrid).* 2002;19:57–8.
- Ruy-Díaz García A, González-Herranz J, Ballesta de Vicente F, Rodríguez Ferrol P. Osteoporosis transitoria de la cadera. Revisión de aspectos clínicos y terapéuticos a propósito de un caso. *Rev Esp Cir Osteoart.* 1994;29:163–9.
- Sellami M, Frikha F, Fourati H, Ezzedine M, Hdjji N, Elleuch MH. Algodyostrophy of the lower limbs during pregnancy. *Ann Readapt Med Phys.* 2006;49:178–86.
- Moltó A, Holgado S, Mateo L, Olivé A. Osteoporosis transitoria de cadera y embarazo. *Med Clin (Barc).* 2010;135:678–9.
- Ribes Iborra J, Fargueta Roig I, Martí-Bonmatí L. Diagnóstico precoz de la osteoporosis transitoria de la cadera versus necrosis isquémica de la cabeza femoral. ¿Existen realmente signos diferenciales? *Rev Cir Osteoart.* 1996;31:163–9.
- Vergara-Ferrer A, Cornet-Flores B, Sevillano González L. Osteoporosis transitoria del embarazo complicada con fractura subcapital de cadera: caso clínico y revisión de la literatura. *Rev Ortop Traumatol.* 2011;55:215–9.

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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/ μ l (90% polymorphonuclear cells, 10% lymphocytes). Due to the polymorphonuclear pleiocytosis we performed a magnetic resonance imaging (MRI) scan (Fig. 1), which showed an extrusion affection with extension 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 Lowenstein), 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

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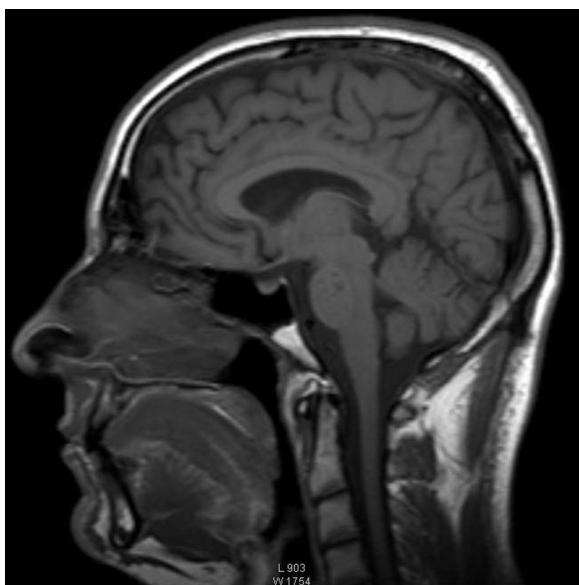


Fig. 1. Brain MRI.

can be seen in early forms of multiple sclerosis and postinfectious encephalopathy. Viral rhomboencephalitis would be a differential diagnosis also, but it is a rare condition in which the injury is unique, extensive and of pontine dominance. Given the history, the clinical and laboratory tests, we oriented the diagnosis toward neuroBehçet.

The patient showed significant clinical improvement and resolution of the headache and nuchal rigidity. A third lumbar puncture was performed 72 h after treatment initiation, with glucose 52 mg/dl, protein 48 mg/dl and 100/ μ l with predominantly mononuclear leukocytes.

Considering the rapid radiographic and clinical improvement after receiving anti-TNF therapy,⁵ we conclude that the patient

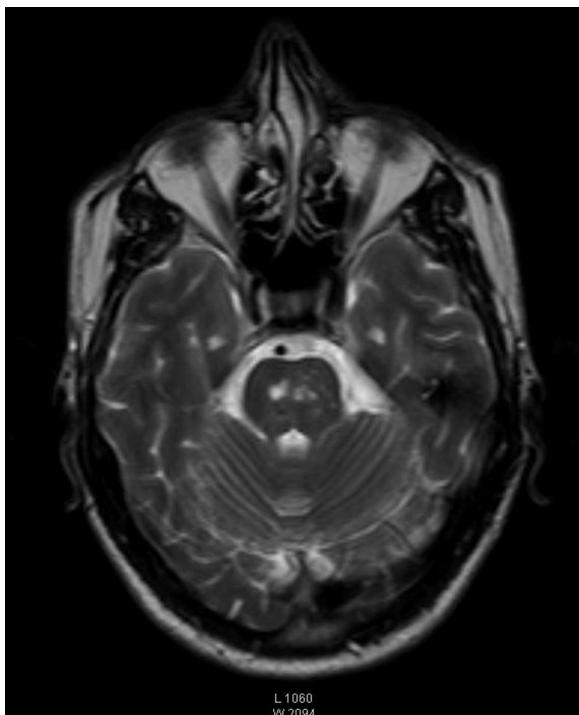


Fig. 2. Involvement of the pons with extension into the left cerebral peduncle.

had BD related aseptic meningitis; the patient was discharged with prednisone 1 mg/kg/day, isoniazide-pyridoxine, infliximab and azathioprine. Control brain MRI showed no lesions described in the previous study and presently the patient had no recurrence of neurological affection.

The central and/or peripheral nervous system affection occurs in about 5%–25% of patients with BD. It is more common in males and usually presents between 3 and 6 years following the onset of illness. Presentation as the first manifestation of the disease is rare.^{6,7}

Involvement of the central nervous system (CNS) is the most prevalent and there are two clinical forms: parenchymal and non-parenchymal affection.⁸ This clinical, pathological, radiological differentiation is important and influences prognosis since parenchymal injury is more prevalent (80% of cases) and has a worse prognosis.⁹ The clinical presentation is nonspecific and varied, including loss of consciousness, seizures, confusion, lethargy, psychiatric disorders, personality changes and dementia.^{10,11} Unlike immunosuppressed patients without BD, meningitis can be present as oligosymptomatic, subacute, or associated to cranial nerve disorders.

In the context of an BD our patient presented neurological manifestations and we established the diagnosis of neuroBehçet supported in patient study and prior exclusion of other causes. The presence of signs of meningeal inflammation is common in the CNS parenchymal form; however, the presentation as isolated meningitis is quite rare. This case highlights, therefore, the importance of ruling out an infectious etiology, particularly in patients undergoing immunomodulator treatment.

MRI findings in relation to the progression of the disease and the initial phase T2 hyperintensity areas in the brainstem, basal ganglia and brain hemispheres, followed by an intermediate stage of edema with a mass effect and microhemorrhages and a third stage with brainstem atrophy have been described.^{12,13} In our case, there was no clinical but rather imaging evidence of thromboencephalic involvement, with findings corresponding in our patient with his initial changes.

In accordance with the above, it is advisable to perform MRI controls for tracking injuries once the treatment is started although these lesions can take months or years to disappear¹⁴; in our case, we verified resolution after 5 weeks of triple therapy with immunomodulatory therapy.

References

- Riera-Mestre A, Martínez-Yelamos S, Martínez-Yelamos A, Ferrer I, Pujol R, Vidaller A. Clinicopathologic features and outcomes of neuro Behçet disease in Spain: a study of 20 patients. *Eur J Int Med.* 2010;21:536-41.
- González G, Pérez R, Satriano R, Rotondo María T. Neuro Behçet: a propósito de un caso clínico. *Arch Pediatr Urug.* 2007;78:139-45.
- Al-Araji A, Kidd DP. Neuro Behçet's disease: epidemiology, clinical characteristics, and management. *Lancet Neurol.* 2009;8:192-204.
- Schwartz RB, Bravo SM, Klufas RA, Hsu L, Barnes PD, Robson CD, et al. Cyclosporin neurotoxicity and its relation to hypertensive neuropathy: CT and MR findings in 16 cases. *Am J Roentgenol.* 1995;165:627-31.
- Pipitone N, Olivieri I, Padula A, D'angelo S, Nigro A, Zucoli G, et al. Infliximab for the treatment of Neuro-Behçet's disease: a case series and review of the literature. *Arthritis Rheum.* 2008;59:285-90.
- Borhani HA, Safari A. Proposing an algorithm for treatment of different manifestations of neuro Behçet's disease. *Clin Rheumatol.* 2010;29:683-6.
- Weiner S, Otte A, Schumacher M, Juengling F, Brink I, Nitsche E, et al. Neuro-Behçet's syndrome in a patient not fulfilling criteria for Behçet's disease: clinical features and value of brain imaging. *Clin Rheumatol.* 2000;19:231-4.
- Latorre González G, Escribano Gascon AB, Lo pez de Silanes de Miguel C, García Cobos R, Ignacio Casanova Peño L, La peña Montero T. Neuro behçet: a propósito de un caso. *Reumatol Clin.* 2009;5:168-70.
- Akman Demir G, Bahar S, Coban O, Tasci B, Serdaroglu P. Cranial MRI in Behçet's disease: 134 examinations of 98 patients. *Neuroradiology.* 2003;45:851-9.
- Atasoy HT, Tunc TO, Unal AE, Emre U, Koca R, Esturk E, et al. Peripheral nervous system involvement in patients with Behçet disease. *Neurologist.* 2007;13:225.

11. Benamour S, Naji T, Alaoui FZ, El Kabli H, El Aidouni S. Neurological involvement in Behcet's disease. 154 cases from a cohort of 925 patients and review of the literature. *Rev Neurol (Paris)*. 2006;162:1084.
12. Akman-Demir G, Serdaroglu G, Taşçı B. Clinical patterns of neurological involvement in Behcet's disease: evaluation of 200 patient. *Brain*. 1999;122:2171–81.
13. Lee SH, Yoon PH, Park SJ, Kim DI. MRI findings in Neuro Behcet's disease. *Clin Radiol*. 2001;56:485–94.
14. Sener RN. Neuro Behcet's disease: diffusion MR imaging and proton MR spectroscopy. *AJNR: Am J Neuroradiol*. 2003;24:1612–4.

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Septic Arthritis Associated to Gout and Pseudogout: The Importance of Arthrocentesis[☆]

Artritis séptica asociada a gota y seudogota: la importancia de la artrocentesis

Dear Editor,

Septic arthritis is a condition that is caused by bacterial nesting in the synovial membrane leading to an inflammatory response that causes the acute purulent synovial fluid appearance. It is a medical emergency because of rapid anatomical and functional impairment. From the etiological viewpoint, Gram-positive bacteria such as *Staphylococcus aureus* (*S. aureus*), are implicated in over 50% of cases, followed by *Streptococcus* (15%–20%) and Gram-negative bacteria. In its epidemiology various risk factors such as advanced age, immunosuppression, presence of prosthetic joints, and patient comorbidities, influence the disease. Sometimes the early diagnosis of septic arthritis is difficult and this increases the risk of joint destruction.¹

We present the case of a patient with two successive episodes of septic arthritis, caused by different microorganisms and micro-crystalline arthritis associated with monosodium urate and calcium pyrophosphate deposits.

The patient, an 86-year-old woman, presented no allergies, no history of substance abuse and was independent for activities of daily living. She had a history of atrial fibrillation treated with warfarin and tophaceous gout. In the last month she presented an episode of septic arthritis of the right shoulder caused by *Escherichia coli* (*E. coli*), due to bacteremia secondary to a urinary tract infection. She was treated with intravenous ciprofloxacin 400 mg every 12 h for 2 weeks (continued with a dose of 750 mg orally for 6 weeks) presenting improvement. Fifteen days after this episode, she presented with pain and swelling of her left knee which had lasted for a week without fever or other symptoms.

On physical examination, the patient was afebrile and stressed left knee monoarthritis with functional impotence for knee flexion. The rest of the examination found no significant alterations. Arthrocentesis was performed, yielding 4 cm³ of liquid of inflammatory characteristics: 9650 leukocytes/mm³ with a predominance of polymorphonuclear cells (94%) and glucose 75 mg/dl. Monosodium urate crystals were observed under polarized light microscopy. Gram stain was negative. The synovial fluid culture was isolated *Escherichia faecium* (*E. faecium*) sensitive only to vancomycin. Laboratory tests showed no leukocytosis (8600/109 leukocytes, 60% neutrophils, 20% lymphocytes) but elevated acute phase reactants (ESR: 100 mm in the first hour, C-reactive protein: 238 mg/dl). Urine

sediment was normal and blood and urine cultures negative. We completed the study with a chest X-ray and echocardiography to rule out pulmonary and/or cardiac affection. A simple X-ray of the left knee showed signs of advanced degenerative joint disease with marked tibiofemoral joint space narrowing and increased soft tissue density and joint effusion. Magnetic resonance imaging showed a tophi in the patellar insertion of the quadriceps tendon that caused a major erosion in the upper pole of the patella (Fig. 1). Antibiotic treatment was initiated with vancomycin, 1 g every 48 h (adjusted for renal function).

Joint lavage was performed daily with saline but was unproductive. After 10 days of antibiotic treatment and the continued signs of knee arthritis, as well as the isolation of the germ in serial cultures, a surgical arthrotomy and debridement with subtotal synovectomy was performed.

The breakdown of the articular surface and the large amount of tophaceous deposits were notable. Cultures after surgery were negative. The pathology of the synovial fluid showed urate and calcium pyrophosphate crystals. The patient remained hospitalized up to a month for intravenous treatment and rehabilitation, which was started early.

Enterococci are Gram-positive diplococci that are part of the normal flora of the human gastrointestinal tract and genital tract of women. They may also be isolated in soil, food, water, plants, birds, insects and other animals. The frequency of isolation of different species varies with the host. *E. faecalis* and *E. faecium* are the dominant species in the human gut, and between the 2 they make up 95% of the microorganisms in the gastrointestinal tract.² Multiple *Enterococcus* cause nosocomial infections (urinary infections, endocarditis, diverticulitis, meningitis and bacteremia) and are resistant to multiple antibiotics. The immunosuppressed population and patients with chronic diseases such as gout are more

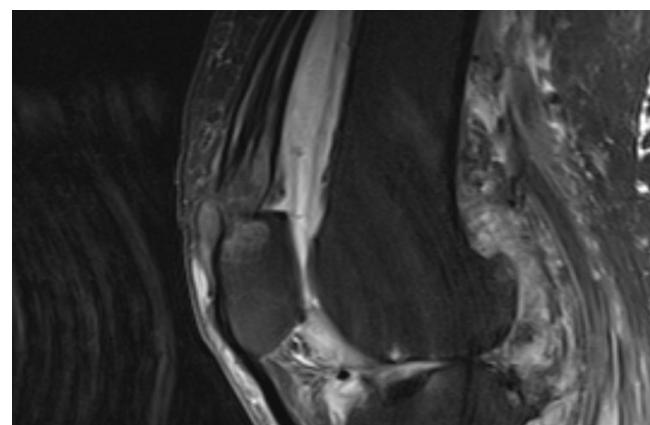


Fig. 1. Hypotensive T2 tophi in the insertion of the quadriceps, causing bone erosion.

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