Acute Meningitis in Behçet’s Disease

Meningitis aguda en la enfermedad de Behçet

Dear Editor,

Behçet’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 neuroBehçet’s, characterized in most cases by the presence of aseptic meningoitis, 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 neuroBehçet 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 pleocitosis we performed a magnetic resonance imaging (MRI) scan (Fig. 1), which showed an extrusion affection with extension to the left cerebral peduncle which appeared isoointense 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 Lownestin), 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 neuroBehçet 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

References

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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.5,7

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

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

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

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.

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 (54%) 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 arthroscopy 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. 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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