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Letter to the Editor

Tolosa-Hunt syndrome as an initial presentation of sarcoidosis☆

Síndrome de Tolosa-Hunt como una presentación inicial de sarcoidosis

Dear Editor,

Tolosa-Hunt syndrome (THS) is defined as an idiopathic granulomatous inflammation of the cavernous sinus or superior orbital fissure, characterised by unilateral painful ophthalmoplegia with abnormalities on brain magnetic resonance imaging (MRI) consisting of an increase in size and hyper-uptake of the intravenous contrast medium in the affected cavernous sinus.[1,2] It usually presents as a painful paralysis of the third cranial nerve, although it can affect any nerve of the cavernous sinus.[1] Diagnosis is clinical and exclusionary. We present a case of THS as an initial presentation of sarcoidosis. This is a 55-year-old woman who consulted with a history of variable, intermittent, horizontal diplopia associated with right upper eyelid ptosis and mild pain. The symptoms had started progressively 4 years earlier and were diagnosed initially as orbital inflammatory disease. The patient also reported erythema nodosum 8 years earlier, and mild fatigue for several months. Physical examination showed paresis of the 6th and 3rd right cranial nerves, with ptosis and horizontal diplopia. In addition, hypaesthesia and allodynia in the territory of the first trigeminal branch (V1). The fundus of the eye was normal bilaterally. Cardiopulmonary, abdominal, skin and locomotor examination were within normal limits. In the complementary tests, blood count, biochemistry, systematic and urine sediment were normal, as well as ESR, PCR and angiotensin converting enzyme (ACE). Lumbar puncture showed normal opening pressure. analysis of cerebrospinal fluid showed no cells, and glucose, total protein and ACE levels were normal. MRI was performed showing swelling and increased

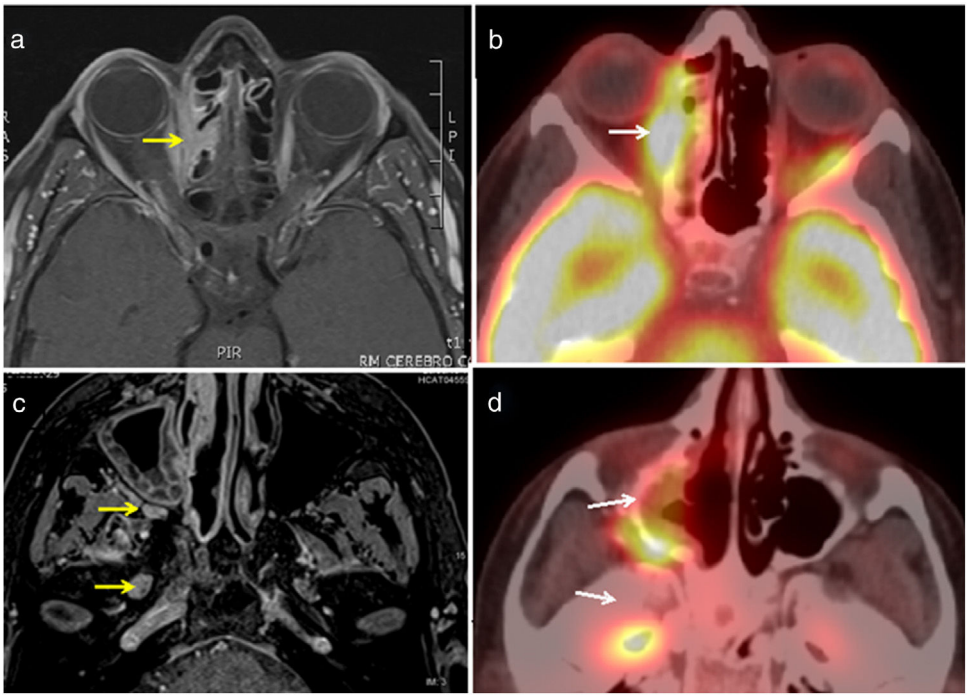

Fig. 1. T1-enhanced axial MRI images with fat suppression after IV contrast (a and c) and PET-CT (b and d): hypermetabolism of the right infiltrative orbital-cranial lesion (extraconal space and ethmoid cells, pterygopalatine fossa and foramen ovale).

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extraocular muscles, with intracranial extension and involvement of the cavernous sinus (Fig. 1a and c). CT scan of the chest showed bilateral hilar lymphadenopathy and small, well-defined lung nodes. An ^{18}F fluorodeoxyglucose PET/CT scan (Fig. 1b and d) showed normal uptake in the right orbit and the lymph nodes of the chest. An endoscopic biopsy of the endonasal cavernous sinus was performed, in which confluent, non-necrotising granulomas suggestive of sarcoidosis were found. Tuberculosis screening was negative.

Given these findings we started treatment with glucocorticoids, first 3 boluses of intravenous methylprednisolone from 1 g/day and then prednisone at a tapering dose of 1 mg/kg. There was rapid clinical improvement, although the orbital extension and established neurological damage showed less response, demonstrated on imaging tests. Given the response to steroids, immunosuppressants were not started.

In our patient, the orbital MRI findings suggested THS. After systemic examination and biopsy, we considered an unusual initial presentation of systemic sarcoidosis with major neurological symptoms. Isolated neurosarcoidosis is rare, since over 90% of patients also have sarcoidosis in other organs, especially the lungs and mediastinal lymph nodes.[3] Cranial neuropathy is the most common manifestation of neurosarcoidosis.[2] Diagnosing neurosarcoidosis is often difficult, because the clinical manifestations and findings of imaging studies can be mimicked by several other diseases. Brain MRI is the most sensitive diagnostic imaging test.[3] This case highlights that sarcoidosis can present in unusual ways, masking neurological disorders.

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Response to: Current Status of Treatment With Intra-Articular Infiltrations in Juvenile Idiopathic Arthritis☆

Respuesta a: Estado actual del tratamiento con infiltraciones intra-articulares en la artritis idiopática juvenil

Dear Editor,

We read the publication by Nieto-González and Monteagudo[1] in Reumatología Clínicawith interest, where a narrative review is undertaken of the literature in connection with the practice of using intra-articular infiltrations with corticoids (IAIC) in patients with juvenile idiopathic arthritis (JIA). We would like to describe the results of a survey of all of the members of the Spanish Paediatric Rheumatology Society (SERPE) in 2017, in which they were asked about their habitual practice in connection with several aspects of the said technique.

The survey consisted of 10 questions devised by the Scientific Committee of the 2017 National Congress of the said society. The questions refer to the medical speciality of the staff who perform IAIC (rheumatologist, paediatrician, rehabilitation doctor or orthopaedic surgeon); the type of JIA used in the said procedure; indication of analgesia and/or sedation during the process; drug(s) used; details of the IAIC technique (asepsis, washing in saline solution, dilution of the corticoid and others); maximum number of joints infiltrated in a single session; maximum number of IAIC in the same joint in one year; recommendations after infiltration; complications after infiltration; and differences between children according to their age. The "SurveyMonkey" platform was used, which makes it possible to create online surveys, (https://es.surveymonkey.com). This platform interprets the replies to surveys and creates basic descriptive statistics for them.

Eighty-five of the 120 members contacted replied. The survey results showed the heterogeneity of the range of professionals who perform IAIC for JIA, as other authors have reported in the past.[2] The appended table shows the said results. In general, the answers to the survey reflect the absence of recognised treatment guides for this technique, showing the existence of practice that is "based on art" in the hospitals that treat young people and children with rheumatic diseases. A North American study that was also based on a survey reached similar conclusions.[3]

This study has several limitations, the chief one of which may be the fact that only 85 or 70% of 120 members responded to the survey. Additionally, not all of the doctors who treat rheumatic diseases in children in our country belong to the SERPE, although the majority do. On the other hand, one potentially interesting analysis was not carried out: to evaluate whether, within the heterogeneous nature of the replies, they could be found to be more homogeneous if classified according to the speciality of the respondent (rheumatologist, paediatrician, rehabilitator or orthopaedic surgeon).

There are other relevant aspects in connection with IAIC technique which we believe were included in the survey. One of these is the increasing importance of ultrasound scan imaging in paediatric rheumatology, as in recent years it has come to be included as another tool[4] for use when infiltrating several locations that are considered to be "difficult" (the temporomandibular joint,[5] tenosynovitis,[6] the tarsal joint[7]). On the other hand, in all fields of paediatrics sedation-analgesic techniques are being adopted to improve the quality of care for paediatric patients; including IAIC, according to several recent publications.[8–10]

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