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CASE REPORT

Garre's chronic sclerosing osteomyelitis with sacral involvement in a child☆

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Abstract We report the case of a 6-year-old patient who started with abdominal pain and left sciatica, which did not improve after applying symptomatic treatment. A complete analytical and imaging study was performed, which showed a lesion in left S1 corresponding to Garrè's chronic diffuse sclerosing osteomyelitis. The diagnosis was confirmed by biopsy of the lesion. Treatment was established with corticosteroids and anti-inflammatory drugs, obtaining a clinical improvement, although in the follow-up imaging tests 2 years after the onset of the symptoms, the lesion persists but with a significant reduction in its size.

PALABRAS CLAVE
Osteomielitis;
Multifocal;
Esclerosante;
Garrè;
Crónico;
Niños;
Sacro

Osteomielitis esclerosante crónica de Garrè en un niño con afectación sacra

Resumen Presentamos el caso de un paciente de 6 años, que comenzó con dolor abdominal y ciatalgia izquierda sin mejoría después de aplicar un tratamiento sintomático. El estudio analítico completo y de imagen realizado mostró una lesión en S1 izquierda correspondiente a osteomielitis esclerosante crónica de Garrè, confirmando el diagnóstico mediante biopsia de la lesión. Se instauró tratamiento con corticoides y antiinflamatorio, que consiguen una remisión parcial de los síntomas, y en las pruebas de imagen realizadas 2 años después del inicio de los síntomas persiste la lesión, aunque con una disminución significativa de su tamaño.

Case report

We present the case of a 6-year-old patient who reported with occasional fever, no circadian rhythm, and severe and diffuse abdominal pain following an accidental fall.

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The symptoms persisted despite treatment with paracetamol and a soft diet for about 10 days. After this period, we decided to complete the study with an abdominal ultrasound and blood tests, which were normal. After approximately 15 days from the onset of symptoms the patient suffered severe, left, lumbosciatic pain and the abdominal symptoms and fever disappeared. We began treatment with non-steroidal anti-inflammatory drugs (NSAIDs) (oral ibuprofen 70 mg/8 h) and analgesic drugs (rectal metamizole 380 mg/8 h), while continuing with imaging studies. We obtained 2 plain radiographic projections of the lumbosacral region, which were normal. The study was completed with a magnetic resonance imaging (MRI) scan of the lumbosacral region which showed a bone lesion accompanied by bone oedema in the left half of the S1 vertebral body and the ipsilateral sacral wing. These could correspond to sequelae of the trauma, stress fractures or infectious processes, without ruling out other diseases.

Since the lumbosciatic symptoms persisted despite treatment with NSAIDs and analgesics, we decided to start treatment with corticosteroids (methylprednisolone 1 mg/kg/day with gradual withdrawal), which temporarily managed to control the symptoms. Laboratory tests including blood count, biochemistry, coagulation, liver function, C-reactive protein, erythrocyte sedimentation rate, complement components C3 and C4, basal ACTH, bone alkaline phosphatase, serum markers for Epstein–Barr virus, cytomegalovirus, parvovirus, antistreptolysin O antibodies, antinuclear antibodies, anti-Ro and anti-neutrophil cytoplasmic antibodies, B27 human leukocyte antigen, urine amino acids, Mantoux and Rose Bengal staining were all normal or negative.

After 3 months of corticosteroid treatment we repeated the MRI scan, which still showed a small, focal area of residual bone oedema, which had become significantly reduced in size as compared to the previous study. This lesion still appeared in MRI controls performed 1 year later, so we obtained a computed tomography (CT) scan of the lumbosacral region, which showed a sclerotic lesion with periosteal reaction in the left wing of the sacrum. This suggested Garrè's chronic diffuse sclerosing osteomyelitis (CDSO) (Fig. 1).

To confirm the suspected diagnosis we obtained a bone scintigraphy scan and biopsy of the lesion. The bone scintigraphy with technetium-99 bisphosphonate in 3 stages showed late uptake in the left sacroiliac region, thus suggesting a chronic inflammatory process. The biopsy revealed fibrofatty tissue and fragments of bone marrow with fibrous areas, but free of specific bone activity and without neoplastic growth. These histological features in conjunction with the radiographic characteristics led to the diagnosis of CDSO.

Approximately 2 years passed since the appearance of the first symptoms until the definitive diagnosis. During this period the patient suffered recurrent, left sciatic pain, with a variable recurrence ranging from 2 to 6 months and the symptoms lasting for 3–5 weeks in each episode.

The patient continues to exhibit the previously described lesion in periodic MRI controls, without significant changes in its dimensions (Fig. 2).

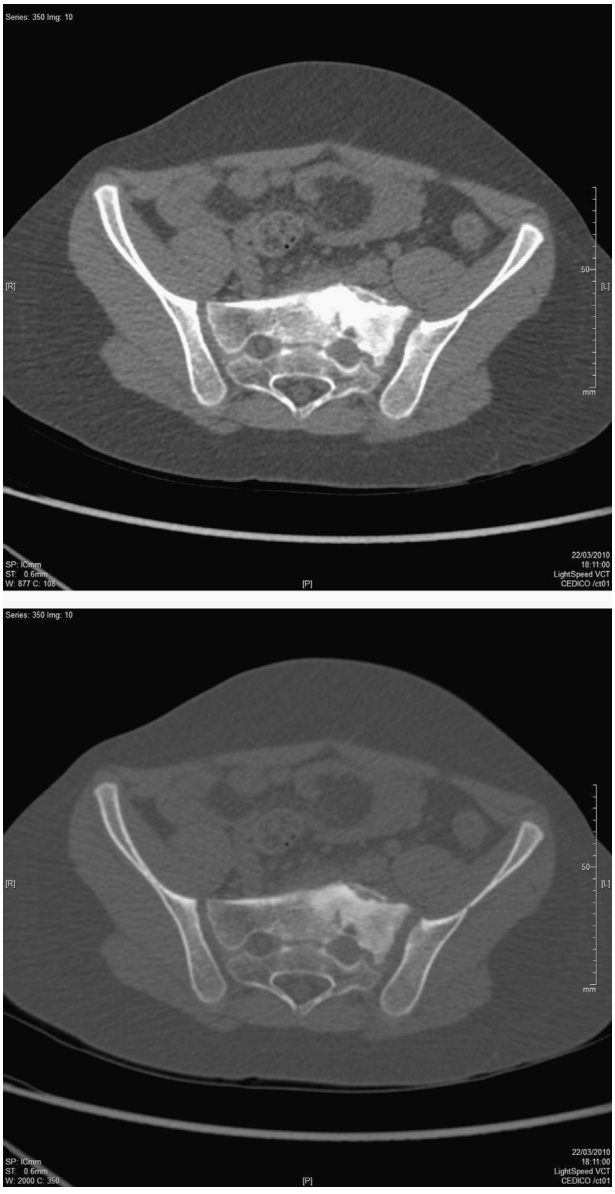

Figure 1 Images of lumbosacral MRI scans in T1, Stir and T2 sequences.

Discussion

CDSO is a type of chronic recurrent multifocal osteomyelitis (CRMO).

It is suspected that CRMO may be an autoimmune disorder. It has been reported in association with several, chronic, autoimmune diseases, including inflammatory bowel disease, Wegener's granulomatosis, psoriasis and Takayasu vasculitis. It is possible that CRMO represents a juvenile form of seronegative spondyloarthropathy[1–6] or a paediatric variant of SAPHO syndrome (synovitis, acne, pustulosis, hyperostosis and osteitis).[7,8]This disease usually appears before the age of 25 years, when the activity of osteoblasts in the periosteum is at its peak.[12] The most common location is the mandible, but it can occur in any bone.[12–15] Most of the patients with CRMO display elevated C-reactive protein or erythrocyte sedimentation rates during exacerbations,