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CASE REPORT

Pyogenic sacroiliitis in the child and young adult. Three case reports

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Abstract Pyogenic sacroiliitis is a rare condition with prevalence of septic arthritis lower than 2% and represents a diagnostic challenge for clinicians due to the variety of clinical expressions, low analytic specificity and the limitations of simple x-rays in the early stages, leading to delayed antibiotic therapy which is the treatment of choice for these patients. The inherent characteristics of its pathophysiology in children makes it distinguishable from that in adults. We report three cases of children and adolescents, two of whom improved clinically after medical treatment, and one required surgical debridement with a postoperative complication.

PALABRAS CLAVE
Sacroileítis séptica;
Sacroileítis piógena;
Resonancia magnética

Sacroileítis piógena en el niño y el adulto joven. A propósito de tres casos

Resumen La sacroileítis piógena es una patología inusual que supone menos del 2%del total de las artritis sépticas y constituye un reto diagnóstico para el clínico debido a la diversidad de manifestaciones clínicas, la baja especificidad de las pruebas analíticas y la limitación de la radiología simple en fases precoces, lo que retarda el establecimiento de una antibioterapia precoz, que ha demostrado ser el tratamiento de elección en estos pacientes. Su diferente fisiopatología en el niño la transforma en una entidad diferenciada de la del adulto. Presentamos tres casos de sacroileítis séptica en la edad infanto-juvenil, de los cuales dos evolucionaron favorablemente con tratamiento médico y uno precisó tratamiento quirúrgico con complicaciones postoperatorias.

Introduction

Pyogenic sacroiliitis is an extremely rare entity, which usually goes unnoticed initially due to a low rate of clinical signs and unspecific symptoms that are not always present, a lack of screening of the sacroiliac (SI) joint and a delay in

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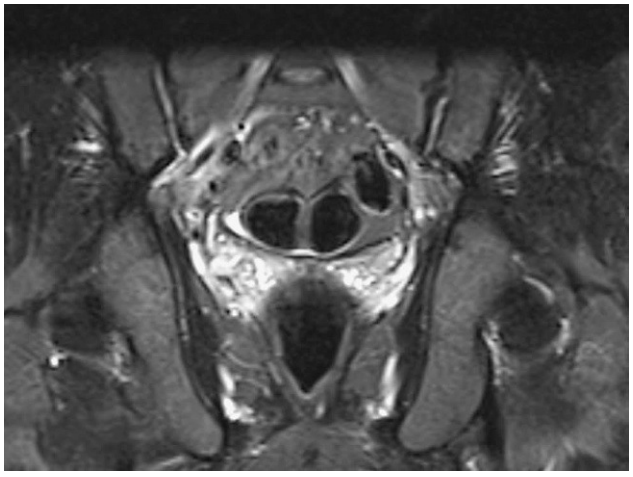

Figure 1 Coronal section of pelvic MRI on T2 STIR sequence on admission, with no pathological findings in either sacroiliac joint.

Figure 2 Sagittal section of pelvic MRI in T2 STIR sequence, 5 days after admission, with increase of the right sacroiliac joint space and adjacent fluid collection in the width of the psoas muscle.

completion of appropriate radiological tests. All of these significantly delay diagnosis in most cases and can give rise to potentially serious complications.

We present three cases of septic sacroiliitis in infantile and juvenile ages, as well as their outcomes in terms of different aetiology, associated morbidity, early diagnosis and therapy. In all cases, the outcome was good, subsequent controls were satisfactory and monitoring was continued until normalisation of laboratory tests in the absence of radiological findings, pain or gait alterations.

Clinical cases

Case 1

Male, 19 years old, with no history of interest, who attended the emergency service due to right coccydynia of a few hours of evolution, with gait difficulty and with no history of trauma.

On admission, he was afebrile and complained of pain in the groin, gluteal region, major trochanter and sacroiliac area, all on the right side. Simple pelvic radiography showed bone immaturity (Risser 4) with no pathological findings. The only abnormal analytical data was the presence of 14,900 leukocytes / mm3 with 91.6% neutrophils. The initial C-reactive protein (CRP) value was around 0.51 mg / dl (normal range 0 to 0.8).

Pain increased during the first 24 hours, with the hip in analgesic flexion, fever peak and CRP value of 4.3 mg / dl. The erythrocyte sedimentation rate (ESR) and procalcitonin values remained normal. Upon suspicion of septic hip arthritis, we requested computed tomography (CT) and magnetic resonance imaging (MRI) scans of the lumbosacral spine and hip, which showed no pathological findings (fig. 1). Abdominal ultrasound was performed and was also normal.

At 48 hours, the case evolved into an established septic condition, with selective pain in the right sacroiliac joint, CRP 22 mg / dl, ESR 43 mm / h (normal range 1-20) and procalcitonin 3 ng / dl (values over 2 indicate risk of sepsis). At this point, antibiotic therapy was started with parenteral amoxicillin-clavulanate until the growth of methicillin-sensitive Staphylococcus aureus in blood cultures, when specific treatment was started with parenteral cloxacillin.

The MRI scan after one week showed oedema and thickening of the gluteal muscles and a thick fluid collection in the right iliopsoas muscle, in an immediately anterior position to the ipsilateral sacroiliac joint (fig. 2). The increase in joint space and T2 signal hyperintensity suggested sacroiliitis as the cause of the abscess.

Debridement of the purulent collection was performed using an ilioinguinal approach. In the immediate postoperative period, there was a large haematoma at the level of the right iliac muscle. This was presumably related to active arterial bleeding points, ruled out by angiography. After clinical stabilisation and normalisation of the acute phase laboratory parameters, the patient was discharged with an oral regime of rifampicin and ciprofloxacin for 4 weeks.

Case 2

Healthy boy, 8 years old, brought to the emergency service with fever of 3 days' evolution accompanied by right coccydynia and progressive functional limitation preventing ambulation. On examination we also noted pain upon pressure in the left renal fossa and SI (painful FABERE manoeuvre, in the absence of Thomas and psoas signs). Laboratory tests showed 15,500 leukocytes / mm3 with 89.4% neutrophils, ESR of 63 mm / h and CRP of 20.6 mg / dl. Initial treatment with anti-inflammatory drugs improved the febrile process, but not the loss of function.

A scintigraphy scan showed increased uptake in right SI (fig. 3) and MRI confirmed the presence of intra-articular

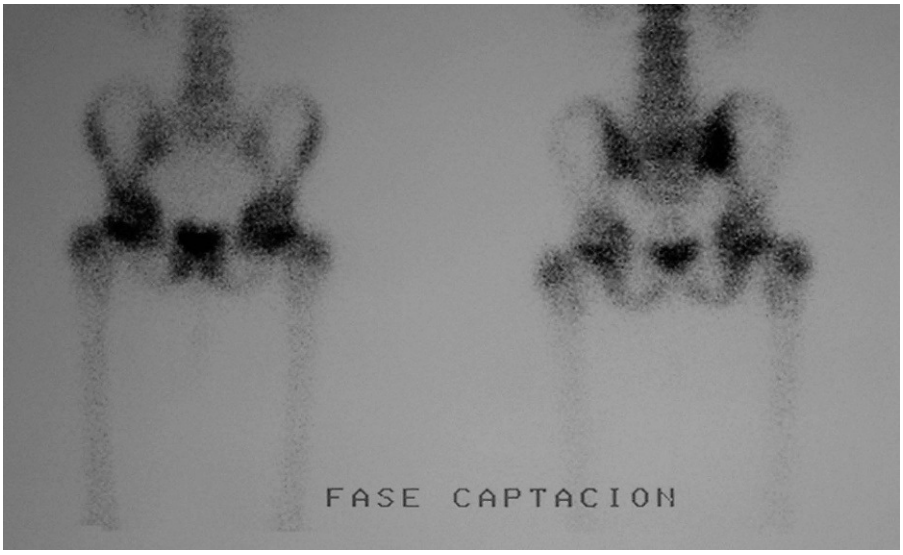

Figure 3 Uptake phase of Tc99 scintigraphy bone scan. The late phase shows an abnormal increased uptake, homogeneous and with notable intensity in the right sacroiliac join; there is also an increased pattern of bone metabolism, consistent with sacroiliitis, with a high degree of inflammatory activity. Quantification by pixels with respect to the sacrum of 1.5 (normally less than 1.3).

fluid (fig. 4), all compatible with right sacroiliitis associated with a small subperiosteal abscess (1 cm in diameter) in the anterior side of the ileum bone, adjacent and lateral to the joint.

Empirical parenteral antibiotic therapy with cloxacillin and cefuroxime was started until a positive blood culture was obtained for *Streptococcus pyogenes*. At this point, the dual therapy was interrupted and cefuroxime was maintained for 18 days until normalisation of CRP and ESR and improvement of pain.

A discrete residual limp while running during the initial days decreased gradually until it disappeared: the patient was discharged from hospital with sequential transfer to oral amoxicillin-clavulanate, which was maintained for 8 weeks, with no trace of the subperiosteal abscess in a control MRI scan.

Figure 4 MRI T2 STIR sequence (1.5 Tesla) of the sacroiliac joint. Increased signal intensity in the ileum and sacrum, adjacent to the right joint and inside the joint, from intra-articular oedema or fluid. Small subperiosteal abscess anterior to the ileum bone, adjacent and lateral to the joint.

Case 3

Healthy boy, 4 years old, without pneumococcal vaccination (PCV7), brought to the hospital with fever, pain in the lower back and gluteus of 24 hours' evolution and inability to walk. He presented an antalgic attitude in prone position and pain in the right SI upon palpation.

The initial laboratory analyses showed an ESR of 31 mm / h (maximum peak, 100 mm / h at 9 days), CRP of 0.8 mg/ dl (5 mg / dl at 2 days) and 20,380 leukocytes / mm3 (78.3% neutrophils).

An ultrasound of both hips (normal) was requested and based on suspicion of septic discitis or sacroiliitis, a pattern of empirical antibiotic therapy with parenteral cloxacillin and cefotaxime was established. An MRI scan of the lumbar spine and both hips revealed oedema in deep fibres of the right iliopsoas muscle and a small leak in the right SI joint, suggestive of inflammation or infection. This was confirmed by an image showing homogeneous pathological increased uptake on a Technetium-99 bone scintigraphy scan. After growth of *Streptococcus pneumoniae* serotype 24 susceptible to penicillin and erythromycin in blood culture, parenteral therapy was begun with cefotaxime and penicillin G for the first days. This was continued until remission of symptoms and normalisation of the analytical results, prescribing oral clindamycin for the following 4 weeks.

Discussion

The sacroiliac joint is involved in pelvic biomechanics, acting as a cushion for the stress caused by changes in load

during ambulation, among others, thanks to the action of the sacroiliac ligaments. These are the most powerful ligaments in the human anatomy, keeping the sacrum in its physiological position within the pelvic ring.[1]

The anatomical characteristics of the joint make it particularly susceptible to infection in certain clinical situations and periods of life. Its vascularization from the pelvic and paravertebral Batson venous systems is highest in young adults, and decreases with age. This situation, associated with decreased joint mobility, reduces the chance of bacterial colonisation in the joint space. Rapid growth stages (adolescence) and hormonal and structural changes taking place during pregnancy increase joint laxity and favour its susceptibility for infectious disease.[2-4]

The SI joint capsule is in contact with the iliopsoas and piriformis muscles in the anterior region and with the gluteal muscles in the posterior region, leading to limping symptoms consistent with irritation of the muscle sheaths and resembling septic arthritis. The anatomical relationship with the lumbosacral plexus and the L5, S1 and S2 nerve roots can also resemble irritation by disc pathology.[2]

Pyogenic sacroiliitis is a rare condition accounting for 1%-2% of septic arthritis cases, usually associated with spondyloarthropathies and other acute inflammatory conditions such as urinary tract infections (up to 40% of cases in some series),[5] pelvic trauma, skin infections, osteomyelitis, septic embolism in endocarditis or other conditions (diabetes mellitus, immunosuppression, RA, HIV type I, prosthetic hip or knee surgery, or direct intervention on the joint).[3,6,7] However, in some series it has not been possible to determine an underlying pathology in up to 40% of cases.[8,9]

The condition can affect all age groups, with a mean age of 20 years and an incidence in patients over 60 years below 3% in some series. There is an even gender distribution in childhood, with a slight predominance of males, whereas in the adult population it is more frequent in women. Children suffer more residual gait alterations compared with adults, in whom atypical presentations, concurrent infections, immune and local complications are more characteristic.[5] It is more often observed in the left sacroiliac joint and has a predilection for the iliac side due to the protection afforded by increased thickness of cartilage on the sacral side of the joint.[10-13]

As in other locations, *S. aureus* is the primary pathogen; with *Streptococcus spp.*, they are the cause of three-quarters of infections. However, there have also been reports of sacroiliitis from *Brucella spp.*, *Mycobacterium tuberculosis*, *Salmonella typhi* and other bacteria.[8,12,14,15]

In two thirds of patients, the onset is insidious, with no typical symptoms (fever, limping and gluteal pain), and presence of delocalized symptoms and radiation to other locations, thus hindering and delaying diagnosis.[11,16] The diagnosis in children may be even more complex because of their lack of cooperation, masked in some cases by an exclusively neurological semiology.[17] Any non-specific lumbago or coccydynia should be associated with a selective screening of the sacroiliac joint using the Patrick or FABER manoeuvres (flexion, abduction, external rotation and extension), Graenslen test (detects a flexion contracture by hyperextension of the affected hip on the edge of the couch and flexion of the contralateral), painful elevation of extended limb, direct compression of the SI or pelvic compression. These manoeuvres are helpful for their sensitivity, specificity and high predictive value, but are rarely performed at the start of symptoms due to a low index of suspicion.[11,18,19]

Blood cultures should be requested before any antibiotic therapy is started, as well as serial analyses including cell count, CRP and ESR.[18]

Standard radiographs are not useful at first, because the changes are only visible in advanced stages and may lead to false negatives, and CT scans are not useful if there is no cortical bone erosion, soft tissue involvement or increased joint space. However, due to the poor result of blood cultures (positive in only 30%-60% of patients), CT is very valuable in obtaining samples for the microbiological study through guided fine-needle aspiration of the joint and, once the abscess is formed, in its delineation and drainage.[2,10,12]

Body scintigraphy with technetium-99 is a highly sensitive test in initial stages, when the pain is poorly localised. It defines the area with increased tracer uptake in the first days after onset of symptoms, but it is not very specific because its low spatial resolution makes it difficult to differentiate an extension caused by sacroiliitis contiguity from an abscess in psoas or gluteus.

An MRI scan is the study of choice because it not only shows joint and marrow involvement, but also the presence of fluid collections, and it provides an early and accurate diagnosis. The scan should include a combination of spin-echo axial and coronal sections, a T1 weighted image (with and without gadolinium injection) and T2, and STIR or fat suppression. Findings include the presence of joint effusion, with a decrease in signal on T1-weighted sequences and an increase on T2 sequences. Inflammatory changes are more evident on T2-weighted fat-suppressed sequences and on T1 with gadolinium injection (Gd-DTPA).[2,18,20] The STIR sequence is the choice for early identification of the extent of infection.[4]

In the adult population, gram-positive germs, especially *S. aureus* and secondly Group A beta-haemolytic streptococcus, are responsible for the majority of septic arthritis cases and 80% of sacroiliitis cases, although the literature reports a considerable number of sacroiliitis by gram-negative germs (*Pseudomonas aeruginosa* and *Escherichia coli*) and anaerobes. Empirical therapy should therefore include bactericidal drugs against all of them.[9] Antibiotic therapy in children and youths should be started with agents against gram-positive bacteria, as these are isolated in the vast majority of reported cases, with predominance of *S. aureus*. In our cases, except for Case 3 (in which double empirical therapy was introduced before the initial suspicion of discitis), the other two were treated with parenteral beta-lactams until the causal germ was isolated. There is currently no consensus and no clinical guidelines have been published on the optimal duration of treatment for septic sacroiliitis. Consequently, although treatment differs according to the causative germ, most authors advocate specific antibiotic therapy in the absence of complications for a

minimum of 4 weeks, or 3 in the case of pneumococcal infection.[11]

Iliopsoas muscle abscess is a rare complication of this entity, which is due to the intimate relationship of the anterior side of the joint with its fascia, but it should always be suspected in the presence of the psoas sign.[21] Major abscesses or sequestering require surgical drainage, usually by a retroperitoneal approach (there have been tests with laparoscopic approaches[10]), although smaller collections may be drained by CT-guided puncture-aspiration to prevent progression towards destructive osteomyelitis, joint destruction and pelvic instability, which may require a subsequent arthrodesis.[22] In our series of 3 patients, Case 1 developed a psoas abscess and surgical debridement was performed due to the magnitude of the purulent collection. However, in Case 3, the subperiosteal location and small size of the abscess did not make its evacuation necessary and radiological tests showed its disappearance at 4 months after symptom onset.

In conclusion, early diagnosis and treatment are crucial in the management of pyogenic sacroiliitis, preventing its evolution towards retroperitoneal abscesses and joint destruction, which increase morbidity and mortality in these patients. Familiarity with this condition, a high index of suspicion and a multidisciplinary approach through collaboration with Emergency, Paediatrics, Internal Medicine, Microbiology, Radiology and Traumatology Services facilitate a favourable evolution in these patients.

Any general examination of patients suffering from lumbago, coccydynia or gait disorders should include selective exercises of the sacroiliac joints to rule out septic sacroiliitis. Specific intravenous antibiotic therapy is the treatment of choice in early stages of the disease. In the absence of complications, this may be sequentially changed to oral therapy as long as it is maintained for at least 4 weeks and follow up is continued until the condition and radiological and laboratory tests return to normal.

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Level of evidence

Level of evidence V.

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