CASE REPORT

Sacroilitis: an unusual cause for a common presentation

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SUMMARY

Inflammatory arthritis and sacroilitis are common presentations to rheumatology clinics. Owing to the physiological changes of pregnancy, the first presentation can be post partum with back pain and an accompanying oligoarthritis or polyarthritis. We present a woman with lower back pain who demonstrated clinical and radiological features consistent with sacroilitis and an inflammatory arthritis but was found to have an unusual presentation of another common cause of arthritis.

BACKGROUND

In the vast majority of patients presenting with oligoarthritis or polyarthritis, this is due to inflammatory arthritis. In the presence of sacroilitis, an inflammatory spondyloarthritis that is usually seronegative to rheumatoid factor and associated with human leucocyte antigen (HLA) B27 should be considered. Very rarely are such symptoms and signs due to diseases such as rheumatoid arthritis, crystal arthritis, connective tissue disease or infection. However, although such causes are rare, given that the treatment for an underlying inflammatory arthritis is immunosuppression it is extremely important that the correct diagnosis is performed. Our case highlights the particular importance of considering uncommon presentations of common causes of acute arthritis in order to avoid potential pitfalls.

CASE PRESENTATION

A 40-year-old woman who was 4 weeks postpartum presented with left hip and lower back pain that had started soon after giving birth. She had a healthy baby through a normal delivery after being induced at 40 weeks but had required an episiotomy. There had been no immediate complications following this. Approximately 12 h after delivery she developed left hip and lower back pain that spread to her left shoulder, wrist and hands associated with a reduced range of movement. Following this she developed pain in her right shoulder and chest pain at the costochondral junctions, and pain and swelling of her right knee. There was no early morning stiffness and no history of psoriasis. She had also noticed ongoing vaginal bleeding since the delivery. She did not report any fevers or rigours.

She had a 16-year history of lower back pain following a 'slipped disc' and a possible radiculopathy. She had been taking diclofenac and co-codamol for a few weeks which alleviated her symptoms. She did

not smoke or drink alcohol and worked as an online editor. There was no family history of inflammatory arthritis nor spondyloarthropathy. There was no recent history of any dental procedures.

Clinical examination showed a subtle swelling around the right sternoclavicular joint and second costochondral junction with significant tenderness. There was a mild reduction in bilateral shoulder movements. Her lower lumbar spine was tender on movement and stressing her right sacroiliac joint provoked pain.

INVESTIGATIONS

Blood tests showed raised inflammatory markers: C reactive protein (CRP) 83 mg/L, (normal <5 mg/L) erythrocyte sedimentation rate (ESR) >120 mm/h (normal 2–12 mm/h). Rheumatoid factor, anticyclic citrullinated peptide antibodies and HLA B27 antigen were negative. She was anaemic: haemoglobin of 7.4 g/dL (normal 11.5–16.5 g/dL), mean corpuscular volume 88 fL (normal 76–96 fL), white cell count $5.6 \times 10^3/\mu\text{L}$ (normal $4.0\text{--}11.0 \times 10^3/\mu\text{L}$), neutrophils $4.4 \times 10^3/\mu\text{L}$ (normal $2.0\text{--}7.5 \times 10^3/\mu\text{L}$), lymphocytes $0.7 \times 10^3/\mu\text{L}$ (normal $1.5\text{--}4.0 \times 10^3/\mu\text{L}$) and platelets $283 \times 10^3/\mu\text{L}$ (normal $150\text{--}400 \times 10^3/\mu\text{L}$). Urea and electrolytes, liver function tests and glucose levels were normal.

A complete MRI was performed along with fat suppression and short tau inversion recovery (STIR) images. This showed abnormal fluid signal within the right sternoclavicular joint space extending into the surrounding tissues, particularly within the interclavicular recess and suprasternal notch (figure 1). The subchondral bone demonstrated

Figure 1 MRI showing abnormal fluid signal in the right sternoclavicular joint and bone oedema.

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Figure 2 MRI showing abnormal fluid in the left sacroiliac joint and bone oedema.

oedema, but no erosions. A large amount of abnormal fluid was present in the left sacroiliac joint extending into the surrounding tissues with pockets of fluid or pus present within the paravertebral muscles and left iliacus muscle. The surrounding bone showed extensive oedema (figure 2).

A nuclear medicine bone scan showed marked increase in uptake at the left sacroiliac joint and the right sternoclavicular joint. There was minimally increased activity in the left shoulder and right knee joint (figure 3). Ultrasound of the small joints of her hands was normal. The features were thought to be suggestive of an inflammatory arthropathy involving the right sternoclavicular and left sacroiliac joint. The diagnosis of exclusion was septic arthritis due to the large amount of fluid in the paravertebral muscles and extensive bony oedema.

Synovial biopsies of her left sacroiliac and right sternoclavicular joint showed small clusters of neutrophil polymorphs suggestive of sepsis and identified a fully sensitive group B *Streptococcus*. The synovial fluid was not sent for crystal analysis as the distribution of arthritis and the clinical background of the patient made crystal arthritis much less likely, especially in view of the positive cultures and biopsies.

An echocardiogram excluded infective endocarditis, with no obvious vegetations. However, it demonstrated a hyperdynamic left ventricle with a right ventricular size at the upper limits of normal with mild pulmonary arterial hypertension.

A gynaecological review revealed retained products of conception, which were evacuated; this was deemed to be the cause of her anaemia. She received a blood transfusion and was started on ferrous fumarate. Following this, the anaemia resolved and did not require any further investigation.

DIFFERENTIAL DIAGNOSIS

The most important diagnosis to consider in patients presenting with oligoarthritis and associated lower back pain is inflammatory arthritis. This includes spondyloarthropathy such as ankylosing spondylitis or psoriatic spondyloarthropathy, which are often associated with the HLA B27 allele. Following pregnancy, inflammatory arthritis can flare up or present for the first time. Rarely, crystal arthritis due to uric acid or calcium pyrophosphate disease can cause spinal involvement, with crystals demonstrated on synovial fluid microscopy. The initial clinical

diagnosis considered in this patient was inflammatory arthritis; however, septic arthritis was high on the differential in view of her clinical presentation. The interesting feature was that she presented initially in a way that was characteristic of an inflammatory spondyloarthropathy. In order to differentiate between inflammatory sacroilitis and infective causes, a thorough history and examination are necessary, along with imaging.

TREATMENT

Our patient was admitted to hospital for intravenous ceftriaxone for 3 weeks, following which she had a 6 week course of oral clindamycin.

OUTCOME AND FOLLOW-UP

There was no clinical or laboratory evidence of an underlying condition such as chronic liver disease, diabetes or long-term immunosuppression that predisposed to development of multifocal septic arthritis. The patient had undergone group B *Streptococcus* screening during her pregnancy with perineal swabs and this had been negative. The baby was healthy and did not have any complications related to the group B *Streptococcus*.

The patient's inflammatory markers improved and her CRP was 29 mg/L (normal <5 mg/L) on discharge. After 12 weeks of antibiotics her CRP was 5 mg/L (normal <5 mg/L) and ESR 11 mm/hr (normal 2–12 mm/h). A repeat echocardiogram was unremarkable with normal pulmonary artery systolic pressure. MRI 6 months later showed a significant improvement of the inflammation previously seen.

In the months following her treatment, she remained well and did not develop any immunocompromising condition such as diabetes mellitus or chronic liver disease that may have been an underlying subclinical cause for the development of her septic arthritis.

DISCUSSION

In recent years, there has been an increase in the rates of adult native joint septic arthritis as well as group B streptococcal (*Streptococcus agalactiae*, or GBS) disease in non-pregnant adults.^{1 2} The emerging antibiotic resistance, particularly to multiple agents, in the responsible organisms can pose challenges to treatment. While septic arthritis classically presents as an acute monoarthritis, it can less commonly present as oligoarthritis, or indeed sacroilitis, as our case illustrates. The most common causative organisms are *Staphylococcus aureus* and *Streptococcus sp.*,³ although the clinical and radiological features are similar. GBS sepsis is widely reported in the literature and although septic sacroilitis is a rare occurrence and difficult to diagnose, GBS is a recognised cause. It tends to occur in younger patients, with the mean age being approximately 20 years and only 3% of cases occurring in patients over the age of 60 years.⁴

GBS is a common cause of maternal and neonatal morbidity and mortality worldwide. Previously a major cause of neonatal sepsis including endocarditis and meningitis, efforts to tackle this have led to a significant decline. It is also responsible for pregnancy-related urinary tract infections, chorioamnionitis and bacteraemia. In the postpartum period, GBS causes significant morbidity and mortality through infections such as mastitis, bacteraemia, meningitis, endometritis and wound infections.⁵ In total there are approximately 50 000 incidences of pregnancy-related infection in the USA each year. However, over the last two decades, there has been a twofold to fourfold increase in the incidence of invasive GBS infections in non-pregnant adults.⁵ Reported rates vary from 4.1 to 7.2 cases/

Figure 3 Nuclear medicine bone scan demonstrating increased uptake in the right sternoclavicular and left sacroiliac joints.

100 000.^{6 7} Skin, soft tissue and articular infections and pneumonia are common and in many cases arise from nosocomial carriage.⁸

The risk of GBS disease is higher in the elderly and those with comorbidities predisposing to invasive infection.^{1 7} The mean age of non-pregnant adults with invasive GBS disease is approximately 60 years. In one study in a large teaching hospital, GBS septic arthritis occurred mainly in elderly patients with predisposing factors being diabetes, malignancy and chronic liver disease.² There is no increased risk of GBS infections associated with recent dental procedures and, indeed, our patient did not have any recent dental history. Most patients with GBS septic arthritis present with monoarthritis. However, in a significant number (32%), more than one joint is involved.^{2 8} The most common joints involved are the knee (36%) and the shoulder (25%).² Most patients also demonstrate a bacteraemia and a concomitant GBS infection in another site such as the urinary tract.

GBS endocarditis can be a major problem in any patient with proven invasive GBS disease. Recent reports suggest it is under-recognised and therefore must be excluded by echocardiography,

as in our patient.⁹ If undetected it can lead to severe valve destruction and also recurrence of invasive GBS disease in other organs. Initially reported in women in the late stages of pregnancy, often with underlying rheumatic heart disease, it is now increasingly prevalent in non-pregnant adults.⁸ The vegetations can be large and friable, leading to large vessel embolic phenomenon. There can also be associated pericarditis, myocarditis, mycotic aneurysms and endophthalmitis.¹⁰

Our patient's multifocal GBS septic arthritis is likely to be a direct result of her pregnancy and episiotomy during labour as GBS are common commensals of the perineum in peripartum women, with a prevalence of approximately 20%. Prolonged rupture of the membranes is associated with increasing colonisation rates, often to over 80%, and a resultant increase in neonatal infection and maternal morbidity.¹¹ In our patient, the Group B *Streptococcus* is likely to have caused infection of the retained products of conception and subsequently haematogenous spread with septic arthritis. Our patient did not present with typical signs of infection such as fever and rigours, in spite of biopsy evidence of multifocal sepsis and had an atypical presentation. Although she had undergone screening prior to giving

birth, this had been negative, suggesting that it had been contracted later. Additionally, there was no evidence of an underlying predisposing condition such as diabetes mellitus or chronic liver disease, nor any other reason to explain the source of her multifocal biopsy positive GBS septic arthritis. Although the evacuated products of conception were not cultured to definitively prove these were the source of infection, there does not appear to be any other likely cause in our patient.

Septic arthritis of the knee and sacroiliac joints has also been described following pregnancy. The bacteria are most likely spread by haematogenous seeding as the internal iliac veins are valveless. Raised intra-abdominal pressure such as that occurring during labour causes blood to be forced through the internal vertebral plexus of Batson via the lateral sacral veins. This may explain the involvement of the sacroiliac joints in pregnancy and post partum.¹¹

CT is useful for defining the extent of bony destruction and for image-guided arthrocentesis.¹² However, MRI is the imaging modality of choice; it allows useful determination of the extent of infection in the sacroiliac joint as well as abscess formation or osteomyelitis. It is more sensitive than CT for bony involvement as it can demonstrate bone marrow oedema, which is often the first sign of osteomyelitis.¹² Anterior and posterior subperiosteal infiltrates and transcapsular infiltrates of the surrounding muscles are characteristic of septic sacroilitis and can be used to differentiate this from spondyloarthropathy.¹² Our patient's MRI scan was in keeping with this as it showed a large left sacroiliac joint effusion extending into the surrounding tissues with pockets of fluid or pus present within the paravertebral muscles and left iliacus muscle in addition to extensive bone oedema. It was the MRI findings that first raised the possibility of a septic arthritis in this patient and led to the synovial biopsies being performed.

Treatment is with appropriate antibiotics: most GBS are sensitive to penicillin with penicillin G being the antibiotic of choice. However, concentrations required for effective clearance of the

bacteria are fourfold to eightfold higher than for group A streptococci.⁸ Additionally, resistance to erythromycin and clindamycin for penicillin allergic patients is increasing and it is importance that allergies as well as sensitivity testing is taken into account when prescribing antibiotic treatment.¹³

Recurrent infections occur in 4.3% of adults and overall mortality of GBS septic arthritis is significant at approximately 25%.⁸ Our case is unusual, as the organism was demonstrated on synovial biopsies from more than one joint. However, prompt diagnosis and correct treatment helped prevent any long-term joint damage and preserved function. Without this, septic joints are often completely destroyed and require replacement,¹⁴ which is far from ideal in a relatively young patient.

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Learning points

- ▶ Although classically an acute monoarthritis, septic arthritis can present less typically as an oligoarthritis or polyarthritis, including, as in this case, a sacroilitis.
- ▶ Early diagnosis and correct treatment with antibiotics are important to prevent irreversible joint destruction and possible joint replacement.
- ▶ In postpartum women, it is important to consider possible causes that may be related to the pregnancy.

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