Acute unilateral sacroiliitis mimicking infection on MRI with response to non-steroidal anti-inflammatory drugs: a distinct presentation of spondyloarthritis?

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Patient consent

All patients in this report have given written consent for publication.

Ethical approval

Institutional review board approval for ethics was not required as patients were managed according to generally accepted standards of care prior to this case series report.

To the Editor,

Sacroiliitis is associated with the spondyloarthropathies (SpA) including ankylosing spondylitis, psoriatic arthritis and reactive arthritis (ReA) and may be visualised using magnetic resonance imaging (MRI). Here, we describe four cases of acute unilateral sacroiliitis with florid MRI appearances that mimicked infection, but demonstrated a prompt and complete response to non-steroidal anti-inflammatory drugs (NSAIDs).

All subjects were HLA-B27 negative males and presented with a rapid symptom onset of unilateral sacroiliitis ranging from 2 days to 4 weeks duration (table 1). One subject had a prior history of ulcerative colitis (case 2) in remission and one had scalp psoriasis (case 3). There were prodromal symptoms in two subjects (cases 1, 4) with short-lived fever at presentation. Case 4 had a sore throat preceding the presentation with neutrophilia ($12.4x10^9$/L) which prompted an infection screen. All four patients demonstrated a significant elevation in acute phase markers with a mean serum C-reactive protein (CRP) of 115 mg/L. There were no overt clinical features of systemic inflammatory response.

Baseline MRI demonstrated florid bone marrow oedema (BMO) in 3 cases (1, 2 and 4) affecting >75% of the sacroiliac joints (SIJ) and moderate (affecting 25-75%) in case 3 (figure 1). High signal was noted in surrounding muscle and soft tissue in all cases by the reporting radiologists who observed the need to exclude infection. Sacroiliac joint aspiration/biopsy was considered in all cases but not conducted due to the prompt symptom response following NSAIDs with improvement in clinical parameters and negative septic screen.

Case 2 was advised to continue empirical combined oral antibiotics for 4 weeks. In addition he continued NSAID therapy for 8 weeks until complete symptom resolution. Case 4 cultured group A *Streptococcus* from a throat swab and borderline anti-streptolysin titre of 466 iU/mL and 406 iU/mL respectively, suggesting plausible post-streptococcal reactive arthritis. Repeat MRI was performed in 3 patients at a mean follow up of 5 weeks which demonstrated improved but persistent inflammatory changes. Further imaging thereafter revealed significant improvement in BMO changes in cases 1 and 3, at 2 and 5 months respectively.

Sacroiliitis typifies SpA but can also occur in sepsis where diffuse soft tissue oedema, in addition to BMO, is characteristic[1]. The symptom onset in SpA can be acute and may include fever and raised CRP, therefore mimicking infection. Bilateral sacroiliitis is invariably inflammatory, however, an acute unilateral presentation is frequently reported in the literature as pyogenic or suspicious for atypical organisms[1]. The current case series demonstrates that acute unilateral sacroiliitis with “extreme”

MRI appearances, particularly with extensive sacroiliac BMO and adjacent periarticular muscle/soft tissue oedema can, despite resembling infection, represent a reactive process suggestive of an inflammatory SpA. These cases illustrate the diagnostic challenge of differentiating infection versus inflammation. This is particularly important given that patients typically present through urgent appointments (cases 3, 4 presented to the emergency department requiring hospitalisation). All patients demonstrated a good response to NSAIDs. Although the dose and duration of NSAIDs required to alter BMO is unclear, our data support previous reports in the literature[2]. We acknowledge that the effect of NSAIDs cannot be quantitatively measured from these series particularly as post-inflammatory changes were still visible in two cases after five weeks. Remarkably, however, all patients were symptom free within eight weeks.

Acute unilateral sacroiliitis can be a manifestation of reactive arthritis[3,4]. During a *Campylobacter jejuni* outbreak, one in fifteen cases of ReA presented with sacroiliitis[4]. Similarly sacroiliitis is a rare manifestation of post-streptococcal reactive arthritis[5]. Pseudo-sepsis has been observed in psoriasis, palmo-plantar pustulosis, acne, and the synovitis acne pustulosis hyperostosis osteitis (SAPHO) syndrome but is an unusual cause for de novo acute unilateral sacroiliitis[6]. The severity of sacroiliitis at baseline regardless of HLA-B27 status, has been shown to be a predictor of poor prognosis for radiographic progression, but little is known specifically for acute reactive arthritis[7]. When managing such cases, it is essential not to overlook infectious sacroiliitis typified on MRI by periarticular muscle oedema, although the cases presented here also demonstrate that inflammatory disease can mimic such appearances[8]. Interestingly and although within the spectrum of SpA, our cases could not be classified according to the ASAS classification criteria given the acute onset of symptoms of less than 3 months duration[9,10].

In conclusion, these case reports highlight that significant reactive inflammatory sacroiliitis can yield MRI appearances mimicking infection. Thorough investigation should always be prioritised but NSAIDs alone can be effective in resolving symptoms over several weeks with eventual patient recovery.

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References
1. Slobodin G, Rimar D, Boulman N, et al. Acute sacroiliitis. *Clin Rheumatol* 2016; 35: 851–856.
2. Varkas G, Jans L, Cypers H, et al. Brief Report: Six-Week Treatment of Axial Spondyloarthritis Patients With an Optimal Dose of Nonsteroidal Antiinflammatory Drugs: Early Response to Treatment in Signal Intensity on Magnetic Resonance Imaging of the Sacroiliac Joints. *Arthritis Rheumatol* 2016; 68: 672–678.
3. Oates JK, Young AC. Sacro-iliitis in Reiter's disease. *Br Med J* 1959; 1: 1013–1015.
4. Timo Hannu K, Kauppi M, Tuomala M, et al. Reactive arthritis following an outbreak of Campylobacter jejuni infection. *J Rheumatol* 2004; 31: 528–530.
5. Mackie SL, Keat A. Poststreptococcal reactive arthritis: what is it and how do we know? *Rheumatology* 2004; 43: 949–954.
6. Inman RD. Conundrums in Nosology : Synovitis , Acne , Pustulosis , Hyperostosis , and Osteitis Syndrome and Spondyloarthritis. 2006; 55: 665–669.
7. Hannu T, Inman R, Granfors K, et al. Reactive arthritis or post-infectious arthritis? *Best Pract Res Clin Rheumatol* 2006; 20: 419–433.
8. Kang Y, Hong SH, Kim JY, et al. Unilateral Sacroiliitis: Differential Diagnosis Between Infectious Sacroiliitis and Spondyloarthritis Based on MRI Findings. *Am J Roentgenol* 2015; 205: 1048–1055.
9. Bennett AN, Marzo-Ortega H, Emery P, et al. Diagnosing axial

spondyloarthropathy. The new Assessment in SpondyloArthritis international Society criteria: MRI entering centre stage. *Ann Rheum Dis* 2009; 68: 765–767.

10. van der Heijde D, Ramiro S, Landewé R, et al. 2016 update of the ASAS-EULAR management recommendations for axial spondyloarthritis. *Ann Rheum Dis* 2017; 76: 978–991.

Table 1.

Clinical characteristics of four HLA-B27 negative subjects presenting with acute unilateral sacroiliitis.

Legend:

* Complete resolution refers to disappearance of symptoms and substantial CRP improvement or normalisation. Yes (Y), No (N), Inflammatory bowel disease (IBD), Ulcerative colitis (UC), Psoriasis (PsO), Uveitis (Uv), Male (M), Female (F), C-reactive protein (CRP), milligrams per litre (mg/L), Erythrocyte sedimentation rate (ESR), millimetres per hour (mm/hr), antibiotics (ABx), White blood cells(WBC), Neutrophils (Neut), Negative (-ve), Blood cultures (BC), microscopy culture and sensitivity (MC&S), Transthoracic echocardiogram (TTE), anti-streptolysin O Titre (ASOT), Group A Streptococcus (Gp. A strep.), Epstein-Barr virus (EBV), cytomegalovirus (CMV), hepatitis B virus (HBV), human immunodeficiency virus (HIV), microscopy culture and sensitivity (M,C&S), antibiotics (ABx) once daily (od), genitourinary (GUM), twice daily (bd), three times daily (tds), four times a day (qds).

Figure 1.

Coronal STIR MRI examination of the sacroiliac joints per case. Cases labelled by corresponding number.

Legend:

Short tau inversion recovery (STIR)