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Septic sacroiliitis: an uncommon septic arthritis

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ABSTRACT

Septic sacroiliitis is an uncommon joint infection and the diagnosis is often delayed. We present the first case of a septic arthritis of the shoulder and of the sacroiliac joint in a woman affected by systemic sclerosis, and we reviewed the medical literature since 1997 to 2008 on septic sacroiliitis with a specific microbiological diagnosis other than Mycobacteria and Brucella species.

Evidence shows that antibiotic therapy should be continued until full clinical and radiological resolution is achieved.

Introduction

Systemic sclerosis is a clinically heterogeneous systemic disorder which affects the connective tissue of the skin, internal organs (such as gastrointestinal tract, lungs, heart and kidney) and, the walls of blood vessels. It is characterised by alterations of the microvasculature, disturbances of the immune system and by massive deposition of collagen and other matrix substances in the connective tissue.

Patients may also exhibit arthritis, which represents the initial symptom in two-thirds of the scleroderma patients, whilst the appearance of inflammatory or septic sacroiliitis has never been described (1).

Septic sacroiliitis (SeSI) is an uncommon osteoarticular infection (1-2 cases reported/year) with often vague symptoms mimicking common conditions such as protruded disk, muscular strain, or visceral pain. Therefore the diagnosis is often missed or delayed. It is typically seen in children and young adults and is rare in middle-aged people especially in those affected by rheumatic diseases (2). In connective tissue diseases all infections are important causes of morbidity and mortality. This increased risk is the result of immune abnormalities and of organ system manifestations associated with these diseases and their treatments (3). Within rheumatic diseases no case of SeSI in scleroderma patients has been described.

In adults the most common predisposing factors for infectious sacroiliitis are intravenous drug use, pregnancy, trauma, endocarditis, hemoglobinopathy, immunocompromised states and infections of the skin and respiratory and genitourinary tracts, but 44% of the cases have no predisposing or associated factors identified. Positive blood cultures have been demonstrated in fewer than 25% of the cases (4). The commonest infecting organism in the general population is *Staphylococcus aureus* (*S. aureus*) (3).

We describe the rare case of a concomitant septic arthritis of the shoulder and sacroiliac joint in an adult affected by systemic sclerosis. Additionally, we reviewed the literature focusing on SeSI with a specific microbiological diagnosis other than Mycobacteria and Brucella species.

Case presentation

A 63-year-old Caucasian woman was diagnosed to have systemic sclerosis, according to ACR 1980 criteria, with diffuse skin and joint involvement in April 2005. She was treated with intravenous prostacyclin analogue (iloprost) infusion. In June 2005 the patient presented an arthritis flare and prednisone therapy was started (0.5 mg/kg for one month, then 0.25 mg/kg for two months and 0.1 m/kg for one more month thereafter).

In November 2005 she was admitted to our Rheumatology Division with a 7-day history of acute low-back pain, progressively worsening, fever and chills. She had no history of trauma and there was no significant personal or family clinical history. During the last iloprost infusion, ten days before her hospitalisation, she had a superficial phlebitis that resolved in a few days with a topical cream not better specified.

On admission she was pyrexial at 39.5°C. Physical examination revealed arthritis of the right shoulder and wrists, tenderness over the right sacroiliac joint. Chest, abdominal, and neurological examination were normal.

The erythrocyte sedimentation rate (ESR) was 74 mm/h, C-reactive protein (CRP) was 735 mg/l (normal value, <5 mg/l), white blood cell count of 11.2 x 10^9/l.

On magnetic resonance (MR) of the right shoulder, T1-weighted images on sagittal plane showed a disomogeneous enhancement due to articular and subacromion bursae synovitis. The pelvic

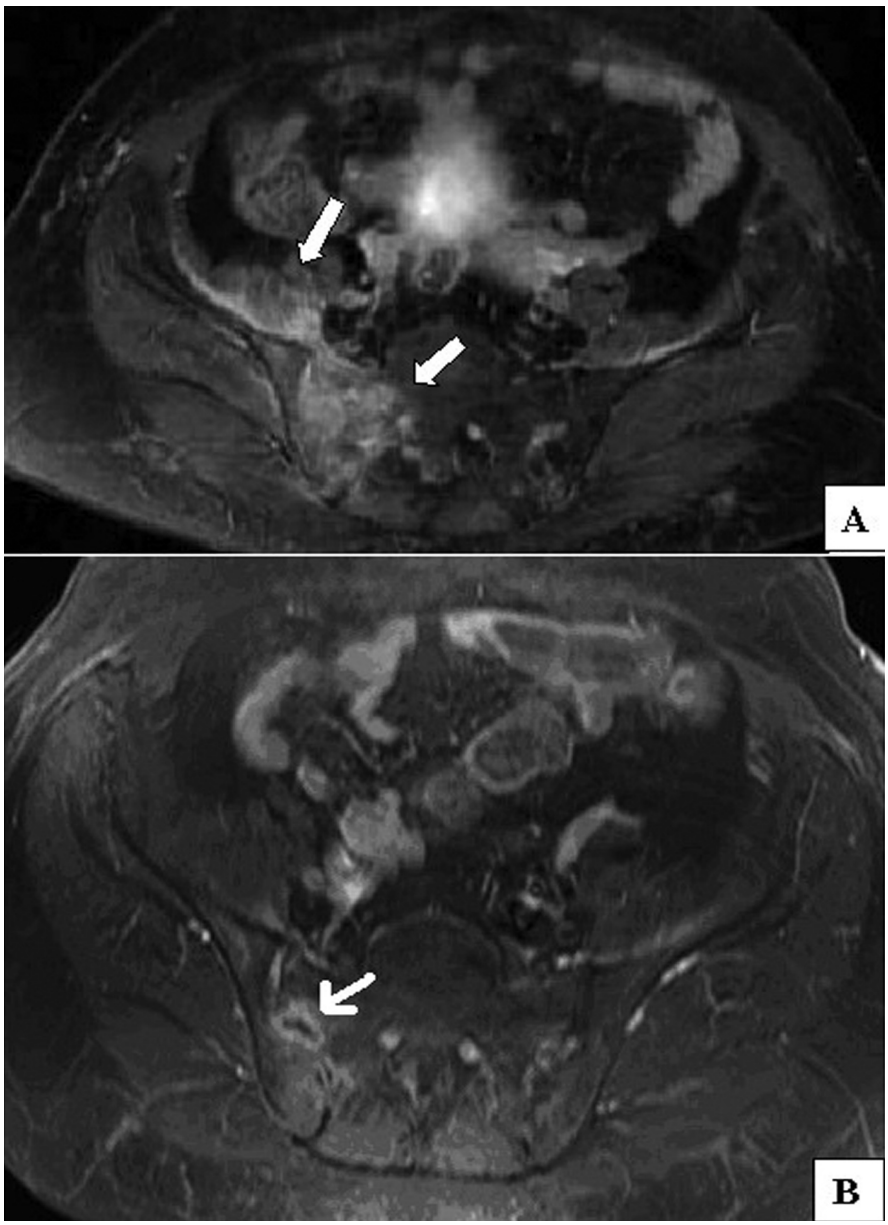

Fig. 1. MRI pelvis.
A. Images T1-weighted fat-saturation post contrast medium. The first examination of the right sacroiliac joint reveals abscessual collections in soft tissues and bone destruction. **B**. After 12 months MR examination reveals only a ring enhancement in the middle of the right sacroiliac synchondrosis.

MR imaging revealed advanced right sacroiliitis with abscessual collections (Fig. 1.A).

A small amount of purulent material from shoulder arthrocentesis was obtained thus confirming the clinical suspicion of septic arthritis. An empirical regimen with intravenous teicoplanin (200 mg/day) was started. Tuberculin skin test, polymerase chain reaction for *Mycobacterium tuberculosis* genome, and serological tests for Salmonella and Brucella were negative. Blood and synovial fluid cultures disclosed *S. aureus*, sensitive to teicoplanin, that was continued.

After 72 hours, the patient had no fever but because of the recovery from the vesical catheter culture of *Enterococcus faecium* resistant to teicoplanin and with intermediate sensitivity to tetracyclines, she also started doxycycline (100 mg twice day per os). This treatment was continued for six weeks; then the patient was discharged with oral doxycycline therapy, prednisone (7.5 mg/day), paracetamol (when needed), and she started the rehabilitation programme.

On dimission, the inflammatory markers were ESR of 56 mm/h, CRP of 88 mg/l. After three months of antibiotic therapy the patient's symptoms improved but did not completely resolve. Because of arthritis in her hands, sulfasalazine (3 g/day) was added to therapy and prednisone tapered (5 mg/day).

At the six-month follow-up, our patient still presented symptoms and radiological signs of active sacroiliitis. therefore we recommended continuing the antibiotic and sulfasalazine therapy until the next check up.

In December 2006, the patient presented scabies and diarrhoea caused by *Candida albicans* (treated with clobetasol and fluconazole with resolution). At the 12-month follow-up, the repeated MR highlighted the sequelae of the septic right sacroiliitis with erosive changes and cavitations (Fig. 1B). The clinical course of patient was good and the antibiotic treatment was stopped. She is continuing the recommended therapy with sulfasalazine.

Review of the literature

Pyogenic sacroiliitis is a relatively rare disorder (1-2 cases reported/year), usually unilateral, which unrecognised (because of its non-specific presentation) or inadequately treated may cause long-term morbidity and eventually lasting disability. SeSI should be considered in the cases of acute or subacute onset of pain in the gluteal region, hip or low back accompanied by fever. It often mimicks features of septic arthritis of the hip, lumbar disc herniation, muscular strain, osteitis of the ilium, or visceral pain and therefore presents a diagnostic rather than a therapeutic problem because the timely administration of antibiotics usually leads to restitution without surgical intervention (5). Laboratory findings are variable and non-specific. In the appropriate clinical setting, imaging techniques such as x-ray computed tomography and MR help to prevent the delay in diagnosis (6).

The most comprehensive reviews of SeSI identified 166 (4) and 177 (2) cases respectively, the first one between 1878 and 1990 and the second one between 1990 and 1996.

Table I. Cases of septic sacroiliitis since 1997 to 2008.

Microorganism (Culture site)	Year	Reference	n.	Age/Sex	Associated condition
Group A *streptococcus*					
(blood)	1998	20	1	24/F	Pregnancy, endocarditis
(blood	2001	9	1	37/F	Post partum
Group B *streptococcus*					
(blood)	1998	18	1	36/F	Curettage
(blood)	1999	22	1	26/F	Pregnancy
(guided joint biopsy)	2000	6	1	28/F	Post partum
(blood)	2001	10	1	36/F	Upper respiratory tract infection
(blood)	2005	11	1	66/M	None
#	2007	37	3	#	#
Str. pneumoniae (blood)	1998	12	1	– /M	Esplenectomia
(blood, cerebrospinal fluid)	1997	13	1	31/F	None
(synovial fluid)	1998	14	1	47/F	Upper respiratory tract infection
(blood, guided joint biopsy)	2001	15	1	62/M	Upper respiratory tract infection
(blood)	2008	16	1	4/M	None
Streptococcus spp. (guided joint biopsy)	2000	6	1	41/F	Suppurative tonsillitis
#	2007	37	1	#	#
S. aureus (blood, perc.needle asp.)	1999	17	1	36/F	Drug use – endocarditis
(blood)	1999	17	1	39/M	Drug use – HIV
(blood)	2001	7	1	63/M	Rheumatoid arthritis
(blood, pus)	2004	19	1	12/F	Trauma
(blood)	2003	5	4	*	None
(blood)	2004	21	1	19/F	None
(blood, synovial fluid)	2001	8	1	55/F	Rheumatoid arthritis
(blood)	2000	6	1	12/F	Trauma
(blood, guided joint biopsy)	2000	6	1	39/F	Unknown autoimmune deficiency
(guided joint biopsy)	2000	6	1	58/M	Deep felon
(guided joint biopsy)	2000	6	1	25/F	Drug use
(guided joint biopsy)	2000	6	1	27/F	Drug use
(blood)	2006	23	1	13/M	None
(blood)	2006	23	1	11/M	None
(blood)	2006	23	1	14/M	None
(blood)	2006	23	1	16/M	None
(guided joint biopsy)	2003	24	1	26/F	Pregnancy
(blood, synovial fluid)	2007	25	1	36/F	None
(synovial fluid)	2007	26	1	30/F	None
#	2007	37	15	#	#
(blood)	2008	38	1	12/M	None
(blood)	2008	38	1	13/F	None
(blood)	2008	38	1	15/M	None
(blood)	2008	39	1	29/F	Upper respiratory tract infection
Enterococcus faecalis (blood, urine)	2000	6	1	10/M	Urinary tract infection
P. aeruginosa (synovial fluid)	2002	27	1	33/M	None
(synovial fluid)	2004	28	1	61/M	Drug use, DM, Tumour
Salmonella spp. (synovial fluid)	2003	29	1	16/F	None
(stool, synovial fluid)	2006	30	1	14/F	None
(synovial fluid)	2001	31	1	15/M	NS
(blood)	2002	32	1	2,5/F	NS
(stool, synovial)	2004	21	1	14/F	NS
(blood)	2007	33	1	31/M	Sickle cell disease (SCD)
#	2007	37	2	#	#
Acinetobacter baumanii	2007	37	1	#	#
Citrobacter freundii	2007	37	1	#	#
NI (synovial fluid)	1999	34	1	26/F	Pregnancy, endometritis
NI (blood culture)	2000	6	1	11/F	None
NI (blood culture)	2000	6	1	11/F	Suppurative tonsillitis
NI (blood culture)	2000	6	1	11/F	None
NI (blood culture, synovial fluid)	2003	5	5	*	None
NI (blood culture)	2007	35	1	40/F	Sacoiliac joint osteomyelitis
NI (blood culture)	2007	36	1	14/F	None
NI (blood culture)	2007	37	10	#	#
NI (blood culture)	2008	38	1	4/M	None
NI (blood culture)	2008	38	1	9/M	None
NI (blood culture)	2008	38	1	12/F	None
NI (blood culture)	2008	38	1	12/F	None
NI (blood culture)	2008	38	1	13/M	None

n: number of cases; F: female sex; M: male sex; NS: not specified; NI: not identified.
* the authors described 6 females and 3 males with a mean age of 27.3 years; 4/6 patients had a positive blood culture
# the authors described 33 patients with a mean age of 36.9 years, 19 females and 14 males; 19/33 patients had a positive blood culture; 9/12 patients had a positive synovial fluid culture

Table II. Differences between demographic and microbiologic characteristics of septic sacroiliitis over time (exclusive of Mycobacteria and Brucella).

	Zimmermann B. 3rd	Our review
Number of cases, n	177	97
Mean age, (range)	20.4 (0.75–72)	29.8 (2,5-80)
Female, n (%)	> M	59 (61)
Age <16 yrs, n (%)	47 (27)	39 (41)
	All cases until 1996	All cases from 1997
Gram-positive microorganisms, n (%)	264 (81)	59 (61)
Species		
Staphylococcus aureus, n (%)	229 (86.7)	41 (69)
Staphylococcus epidermidis, n (%)	5 (1.9)	–
Streptococci, n (%)	29 (11)	17 (29)
Enterococcus faecalis, n (%)	1 (0.4)	1 (2)
Gram-negative microorganisms, n (%)	55 (16.9)	12 (13)
Species		
Pseudomonas aeruginosa, n (%)	17 (31)	2 (17)
Salmonella species, n (%)	16 (29.1)	8 (67)
Escherichia coli, n (%)	8 (14.5)	–
Enterobacteriacae spp, n (%)	3 (5.5)	1 (8)
Neisseria species, n (%)	7 (12.7)	–
Haemofilus species, n (%)	2 (3.6)	–
Altri, n (%)	2 (3.6)	1 (8)
Gram-variable organisms, n (%)	2 (0.6%)	–
Anaerobic microorganisms, n (%)	4 (1.2%)	–
Fungus, n (%)	1 (0.3%)	–
No growth, n (%)	–	26 (27)
Total of cases, n	326	97

M: male sex.

We did an extensive PubMed electronic database search in the English and foreign language medical literature citations with the key words "septic sacroiliitis" from 1997 to 2008 and 97 reported cases of SeSI (Mycobacteria and Brucella cases excluded) (Table I and II) were identified. The mean age of the patients at the onset of infection was 29.8 years (range 2.5-80 years). There was a slight predominance of females (59 of 97 patients, 61%). Paediatric cases accounted for 39 patients (41%). Among rheumatic diseases only two cases in rheumatoid arthritis patients are described (7, 8). Most cases are caused by gram-positive microorganisms (59 of 97 cases, 61%) of which *S. aureus* was by far the most common (69%), confirming previous data published by Zimmermann 3rd (87%). Among gram-negative bacteria (12 cases), our review highlights an increased frequency of *Salmonella species* sacroiliitis (67%), compared to what evidenced by Zimmermann 3rd (29%). Indeed, his review revealed *Pseudomonas aeruginosa* as the most common gram-negative organism that's was seen almost exclusively in immunocompromised, hospitalised subjects or intravenous drug abusers. In 26 (27%) cases no etiologic organisms has been identified from cultures of biological samples.

Blood cultures were performed in 80 of 97 patients and 55 patients grew organisms (69%).

The treatment of SeSI has not been standardised in the literature. Nevertheless, the majority of the authors suggest a minimum of 2 weeks of intravenous antibiotics followed by 6 weeks of oral antibiotics (5); according to other authors, prolonged antibiotic treatment for 46 weeks is recommended (7). Imaging is useful during follow-up of these patients, considerating that clinical improvement precedes resolution of the MR imaging findings. The MR features were seen to persist for a long time despite clinical resolution; the regression on MR imaging was seen only after a prolonged course of antibiotic therapy (6).

Discussion

SeSI is a very rare infection especially among rheumatic diseases and no case in scleroderma patients have been described up to now. To the best of our knowledge, our case is the first one reported in course of systemic sclerosis and again it stresses the likely predisposing role played by glucocorticoids treatment in a patient whose the underlying disease state may have contributed to the development of infectious complication. Healthy humans are carriers (on their skin) of *S. aureus* without any active infection or disease. *S. aureus* infections are common in people with frequent skin injury, particularly if the skin is dry. Wherever skin tissue is damaged such locations provide even better opportunities for *S. aureus* to attach and colonise. Therefore, we suppose that this severe and rare septic complication was triggered by the recent superficial phlebitis (injury on the dry and sclerotic skin that the patient had 3 days before the onset of symptoms during the iloprost infusion by peripheral venous catheter placed in her forearm) in a subject with a relatively immunocompromised state secondary to corticosteroid therapy.

Although it is hard to accept the relatively immunosuppression state induced by steroids as a predisposing cause because of the small amount and brief duration of therapy, it remains that the patient had no other known causative factors for pyogenic sacroiliitis (intravenous drug use, pregnancy – she was in menopause – trauma, endocarditis, hemoglobinopathy, infections of the skin, respiratory or genitourinary tracts), apart from the phlebitis that may have been the trauma and facilitated the S. aureus colonisation. Atzeni *et al*., focused the risk of infections in patients with rheumatic diseases due to various anti-rheumatic drugs, among them the glucocorticoids. In a prospective cohort study on patients with inflammatory polyarthritis the overall incidence of infection was two and a half times that of the general population and one of the significant independent predictors was the use of glucocorticoids; unfortunately, no data were given concerning the doses of the glucocorticoids (40).

A long-term antibiotic therapy was effective in attaining a restitutio ad integrum, above all the therapeutic association with sulfasalazine. Systemic sclerosis is a connective tissue disease that can involve also the osteoarticular system and patients can complain of arthralgias or arthritis, that often represent the initial symptom of the disease, whilst the appearance of inflammatory or SeSI has never been described. We report the description of the first case and we believe this unusual osteoarticular involvement to be as a direct results of a skin trauma (on the skin already involved by the underlying disease) in a patient with a relative immunosuppression state. In conclusion, SeSI, although remaining a rare event, should remind us of the higher susceptibility of patients with inflammatory diseases to develop joint infections, such as sacroiliitis, an occurrence that presumably is worsened from immunosuppressive treatments.

Moreover, our experience shows that antibiotic therapy should be continued until full clinical and radiological resolution is achieved.

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