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Infectious Sacroiliitis: A Retrospective Analysis of Ten Cases

İnfeksiyöz Sakroileit: On Vaka ile Retrospektif Analiz

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ABSTRACT

Introduction: *Infectious sacroiliitis is a rare disease that can cause severe loss of function in patients. In this study, we aimed to analyze the clinical findings, diagnosis, and treatment of infectious sacroiliitis.*

Materials and Methods: *Ten patients who were admitted to the Mersin City Training and Research Hospital Infectious Diseases and Clinical Microbiology Clinic between February 2017 and February 2022 and diagnosed with infectious sacroiliitis were included in the study. The clinical findings, co-morbidities, laboratory, radiological results, and treatments of the patients were accessed from the patient files and hospital information system.*

Results: *Of the ten cases of sacroiliitis, three were men and seven were women. The average age was 51 years. All patients had complaints of lumbo-gluteal pain and difficulty walking. Four of the ten patients included in the study had a positive Brucella slide agglutination test and a tube agglutination test of 1/160 and above, and these patients had no prior history of Brucella infection. Contrast-enhanced sacroiliac magnetic resonance imaging revealed right sacroiliitis in four patients, left sacroiliitis in three patients, and bilateral sacroiliitis in three patients. The mean complete cure time for magnetic resonance imaging (MRI) was 4.8 months, and the mean effective treatment time was 5.9 months in all cases.*

Conclusion: *In cases of infectious sacroiliitis, the microbiological agent cannot always be isoted. Therefore, treatment may be delayed. As seen in our study, when the specific causative agent cannot be isolated, a diagnosis based on treatment may be helpful in such patients.*

Key Words: *Sacroiliitis; Infectious sacroiliitis; Bone and joint infections*

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ÖZ

İnfeksiyöz Sakroileit: On Vaka ile Retrospektif Analiz

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Giriş: *İnfeksiyöz sakroileit, hastalarda ciddi fonksiyon kaybına neden olabilecek nadir görülen bir hastalıktır. Bu çalışmada, polikliniğimize başvuran on sakroileit olgusuyla beraber, infeksiyöz sakroileitin klinik bulguları, tanı ve tedavisinin irdelenmesi amaçlanmıştır.*

Materyal ve Metod: *Şubat 2017-Şubat 2022 yılları arasında Mersin Şehir Eğitim ve Araştırma Hastanesi İnfeksiyon Hastalıkları ve Klinik Mikrobiyoloji Kliniğine başvuran ve infeksiyöz sakroileit tanısı alan on hasta çalışmaya dahil edildi. Hastaların klinik bulgularına, ek hastalıklarına, laboratuvar, radyolojik sonuçlarına ve tedavilerine hasta dosyaları ve hastane bilgi sisteminden ulaşıldı.*

Bulgular: *On sakroileit vakasının üçü erkek ve yedisi kadındı. Yaş ortalaması 51 yıldı (yaş aralığı= 24-91 yıl). Tüm olguların lumbo-gluteal ağrısı, yürümede zorlanma şikayeti vardı. Çalışmaya alınan on hastanın dördünde Brusella lam agglutinasyon testi pozitif ve tüp agglutinasyon testi 1/160 ve üzerindeydi ve bu hastaların daha önce Brusella infeksiyonu geçirme öyküsü yoktu. Çekilen kontrastlı sakroiliak manyetik rezonans görüntülemelerinde dört hastanın sağ sakroileiti, üç hastanın sol sakroileiti ve üç hastanın bilateral sakroileiti mevcuttu. Tüm olgularda manyetik rezonans görüntülemelerinde tam kür zamanı ortalama 4.8 ay, etkin tedavi süresi ise ortalama 5.9 ay olarak tespit edildi.*

Sonuç: *İnfeksiyöz sakroileit olgularında mikrobiyolojik etkenin tespit edilmesi her zaman mümkün olmamaktadır. Bu nedenle tedavide gecikmeler yaşanabilir. Çalışmamızda da görüldüğü gibi spesifik olarak etkenin tespit edilemediği durumlarda tedaviden tanıya gitmek bu hastalarda yardımcı olabilir.*

Anahtar Kelimeler: *Sakroileit; İnfeksiyöz sakroileit; Kemik ve eklem infeksiyonları*

INTRODUCTION

Sacroiliitis is the inflammation of the sacroiliac joint. Treatment strategies and prognosis vary according to the causes of sacroiliitis. Sacroiliitis may be associated with sacroiliac arthritis, ankylosing spondylitis, osteitis condensans ilii, gout, osteoarthritis, tumors, septic arthritis, and brucellosis[1]. Risk factors for infectious sacroiliitis include trauma, pregnancy, use of intravenous drugs, endocarditis, and immunosuppression. Laboratory values are non-specific, and the most common abnormality is increased inflammatory markers. Magnetic resonance imaging (MRI) is the most sensitive imaging method for the diagnosis of sacroiliitis[2]. Infectious sacroiliitis constitutes 1-4% of bone and joint infections. As with other bone and joint infections, it is often caused by bacteremia but can also be caused by an adjacent infection[3]. The most common clinical symptom has been reported to be lumbo-gluteal pain, but coxofemoral pain, pubalgia, and abdominal pain have also been observed. The presence of fever is variable. The clinical picture may be mistaken for sciatica or spondylodiscitis and may cause delays in diagnosis. The causative organism has been reported to be gram-positive cocci, predominantly *Staphylococcus aureus*. It has been reported that in cases where the causative agent cannot be identified, *S. aureus* should be treated, and in case of treatment failure, antibiotic treatment should be broadened to include gram-negative bacteria[4].

MATERIALS and METHODS

The study included ten patients who were admitted to the Mersin City Training and Research Hospital Infectious Diseases and Clinical Microbiology Clinic between February 2017 and February 2022 and diagnosed with infectious sacroiliitis. The patients were examined by the rheumatology department and were diagnosed with infectious sacroiliitis without rheumatological pathology. Written informed consent forms were obtained from the patients. Patients' clinical findings, comorbidities, laboratory and radiological results, and treatments were retrieved from patient files and the hospital information system. Laboratory evaluation of the patients included C-reactive protein (CRP),

erythrocyte sedimentation rate (ESR), leukocyte count, *Brucella* slide, and tube agglutination test and radiological evaluation was performed with contrast-enhanced sacroiliac MRI. As the patients were followed up as outpatients, hemocultures could not be collected. QuantiFERON-TB GOLD test was not performed as it is not available in our hospital. MRI full cure time was defined as full recovery in contrast-enhanced sacroiliac MRI of patients, and effective treatment time was defined as clinical complete recovery with radiological recovery in patients.

RESULTS

Of the ten cases of sacroiliitis, three were men and seven were women. The average age was 51 years (age range= 24-91 years). Two patients had hypertension and diabetes mellitus. The other eight patients had no comorbidities. Patients had no history of hospitalization or operation in the last year. Patients had no family or personal history of frequent urinary tract infections, soft tissue infections, Brucellosis, or tuberculosis. All patients had complaints of lumbo-gluteal pain and difficulty walking. Three patients (3/10) had high fever and fatigue. No weight loss was noted in the patients. The mean symptom duration was 34 days (14-65 days).

The ratios of symptoms and laboratory values according to the infectious agents are shown in Table 1. PPD was above 10 mm in one patient, below 10 mm in six patients, and anergic in three patients. Contrast-enhanced sacroiliac MRI revealed right sacroiliitis in four patients, left sacroiliitis in three patients, and bilateral sacroiliitis in three patients. In our study, based on MRI findings, T2A signal increases and diffuse contrast enhancement were observed on the right sacroiliac joint surface in four patients. Additionally, in one patient, a slightly hyperintense, contrast-enhancing abscess measuring approximately 7 x 5 mm was detected on the left sacroiliac face. Edematous T2A signal increases in the left sacroiliac joint mid-section anterior and iliac subcortical bone marrow and sacral subcortical bone marrow in two patients and T2A hyperintense signal changes in the synovium in this area, and periarticular mild T2 signal intensity increases were observed in three patients.

Six patients in whom the causative agent could not be identified were empirically treated with teicoplanin IV 1 x 600 mg and ciprofloxacin 2 x 750 mg tablets. Four patients with positive *Brucella* agglutination test of 1/160 and above were started on doxycycline 2 x 100 mg tablets, rifampicin 1 x 600 mg tablets, and streptomycin intramuscular 1 x 1 g for 14 days. In a patient receiving Brucellosis treatment, teicoplanin intravenous 1 x 600 mg and ciprofloxacin 2 x 750 mg tablets were initiated due to the failure to achieve MRI and clinical improvement despite two months of treatment. However, since the patient did not benefit clinically from this treatment for one month and there was no improvement in MRI, anti-tuberculosis (anti-TB) treatment (First two months; INH (isoniazid) 1 x 300 mg tablet,

Table 1. Ratios of symptoms and laboratory values according to the causative agent in cases of infectious sacroiliitis

Symptoms and Laboratory Values	All sacroiliitis cases (n= 10)	Sacroiliitis due to *Brucella* spp. (n= 4)	Sacroiliitis due to other infectious agents (n= 6)
Fever	3 (33.3%)	3 (75%)	0
Lumbo-gluteal pain	10 (100%)	4 (100%)	6 (100%)
Leukocytosis	0	0	0
CRP positivity	5 (50%)	3 (75%)	2 (33.3%)
Procalcitonin positivity	4 (40%)	3 (75%)	1 (16.6%)
ESR positivity	6 (60%)	3 (75%)	3 (50%)
Brucella tube agg. (over 1/160)	4 (40%)	4 (100%)	0

CRP: C-reactive protein, ESR: Erythrocyte sedimentation rate.

rifampicin 1 x 600 mg tablets, ethambutol 1 x 1500 mg tablets, pyrazinamide 1 x 2000 mg tablets, maintenance therapy; INH 1 x 300 mg tablet and rifampicin 1 x 600 mg tablets) was started and the patient benefited clinically from anti-TB treatment and complete cure was observed on MRI with 12 months of anti-TB treatment. The PPD of this patient was anergic. Another patient who was treated for pyogenic infection did not achieve clinical and imaging benefits from two months of teicoplanin intravenous 1 x 600 mg and ciprofloxacin 2 x 750 mg tablets treatment and was from an endemic region in terms of tuberculosis, and was therefore switched to anti-TB treatment (First two months; INH 1 x 300 mg tablet, rifampicin 1 x 600 mg tablets, ethambutol 1 x 1500 mg tablets, pyrazinamide 1 x 2000 mg tablets, maintenance therapy; INH 1 x 300 mg tablet and rifampicin 1 x 600 mg tablets) and achieved complete cure with anti-TB treatment in nine months.

In three patients who started Brucellosis treatment, the mean effective treatment time was five months, and the mean MRI complete cure time was four months. In five patients treated for pyogenic infection, the mean duration of effective treatment was 6.5 months and the mean time to complete MRI cure was 3.1 months. In all cases, the mean time to complete the MRI cure was 4.8 months and the mean duration of effective treatment was 5.9 months. The longer duration of effective treatment compared to the time to achieve a complete MRI cure was suggested to be attributed to the patients' continued medication usage after the follow-up MRI was conducted and their delayed attendance at the follow-up visit.

DISCUSSION

Sacroiliitis is a painful inflammation of the sacroiliac joint which is difficult to diagnose. Inflammation can develop secondary to autoimmunity, microtrauma, exercise, and infections. Sacroiliitis may also be associated with Crohn's disease, inflammatory bowel disease, ulcerative colitis, and gout[5]. Sacroiliac infection is extremely rare, occurring in only 1-4% of bone and joint infections. It is often unilateral but there have been reports of bilateral infections[6]. In a study of 136 cases of sacroiliitis conducted in 2016, the rate of infectious sacroiliitis was 8.8%. The study also reported a rate of 61.8% for bilateral sacroiliitis and 38.2% for unilateral sacroiliitis[1]. In our study, seven (7/10) patients had unilateral sacroiliitis, whereas three (3/10) patients had bilateral sacroiliitis. The most common symptom was pain in the lumbo-gluteal region[4,7]. Lumbo-gluteal pain was noted in all ten cases included in our study. All patients except those with brucellosis had no fever. The most common predisposing factors for infectious sacroiliitis in adults were the use of intravenous drugs, pregnancy, trauma, endocarditis, hemoglobinopathy, immunocompromised conditions, and skin, respiratory, and genitourinary system infections, but no predisposing or associated factor could be identified in 44% of cases[8]. The cases in our study had no serious risk factors for sacroiliitis.

S. aureus is the most common causative agent of infectious sacroiliitis[4,9]. Although gram-positive microorganisms were most frequently observed in the study, an increase in *Salmonella* cases was emphasized. *Pseudomonas aeruginosa* is often seen in immunocompromised, hospitalized patients and intravenous drug addicts[8]. In a study involving adult and pediatric patients, *S. aureus* was the most common causative agent, followed by group B and D streptococci and *Salmonella*[10]. In a case report published in Korea, methicillin-resistant *S. aureus* was identified as the causative agent and reported to be community-acquired[9]. Cases of sacroiliitis due to Brucellosis and tuberculosis as specific causes of sacroiliitis have also been reported[11-13]. In a study presented as a case report, a patient with autoimmune hemolytic anemia and chronic steroid use was reported to have cryptococcal sacroiliitis[14]. As the patients included in our study were followed up as outpatients, hemocultures could not be performed. Therefore, we could not determine the causative agent except for *Brucella* spp. which was serologically diagnosed.

Leukocytosis, positive Wright's test, positive Coombs Wright's test, and positive 2-mercapto ethanol test were significantly more common in patients with infectious causes of sacroiliitis, while increased ESR and CRP levels were more common in patients with non-infectious

inflammatory causes of sacroiliitis[1]. In another study, CRP value was increased in all patients, whereas leukocytosis was observed in only 46.8% of patients[4]. In cases of infectious sacroiliitis, it was found that leukocytosis is not a sensitive marker for the detection of pyogenic sacroiliitis, CRP is more sensitive, ESR and CRP may be elevated in most cases, but although they are sensitive, they are not specific[10,15]. In our study, none of the patients had leukocytosis, five patients had elevated CRP and six patients had elevated ESR. In the radiological diagnosis of sacroiliitis, MRI has been reported to perform better than computed tomography (CT) in the evaluation of soft tissue abscess formation and early detection of bone marrow edema and effusion[16]. Magnetic resonance imaging is the reference imaging method, which also determines whether the infection has spread to adjacent muscle structures[4]. Studies have shown that non-iliac dominant pattern bone marrow edema is more common in infectious sacroiliitis. Large bone erosions have only been found in cases of infectious sacroiliitis[17]. In a review, it was stated that the common MRI features of septic sacroiliitis, joint space, periarticular muscle tissue, and anterior and/or posterior subperiosteal infiltrates are hypointense on T1-weighted images and hyperintense on T2-weighted images[18]. In our study, consistent with the literature, edema, and periarticular signal increases were observed in the sacral and iliac bone marrow. However, large bone erosion, thick capsulitis, and extracapsular fluid collection were not observed. To better understand the differences between infectious and non-infectious sacroiliitis on MRI, it is recommended to conduct studies with larger patient cohorts comprising both infectious and non-infectious sacroiliitis groups. Furthermore, it has been reported that ultrasonography is beneficial in ruling out hip joint effusions, while CT imaging is particularly valuable for identifying bone abnormalities and assisting in the guidance of aspiration or biopsy procedures. Treatment of septic sacroiliitis includes 4-8 weeks of parenteral antibiotic therapy and oral antibiotic therapy to prevent recurrence[19]. In the absence of any identified microorganisms, it is preferable to consider active antibiotic therapy for Staphylococcus; and, in case of failure, antibiotic therapy should be broadened to include gram-negative bacilli[4,10,15]. Specific treatments for tuberculous sacroiliitis include anti-TB treatment for at least twelve months and surgery[20]. Combination treatments of rifampin, doxycycline, and streptomycin/gentamicin are also recommended in *Brucella*-associated sacroiliitis[21].

The limitations of our study were the lack of hemoculture because the patients were followed up as outpatients, the lack of QuantiFERON-TB GOLD test which was not available in our hospital, and the inability to detect the causative agent due to the lack of surgical sampling. However, since *S. aureus* was the most common causative agent of infectious sacroiliitis, treatment was directed against this agent, and a complete cure was achieved in all but one patient. Only one patient did not show improvement with teicoplanin and ciprofloxacin treatment, despite receiving an adequate duration of treatment. Since this patient was from an endemic region for tuberculosis, anti-TB treatment was initiated. The patient demonstrated improvement. In a case suspected to have *Brucella*-associated sacroiliitis, the patient did not benefit from teicoplanin and ciprofloxacin treatment, which was switched due to a lack of clinical and imaging improvement despite treatment for *Brucella*, and a complete cure was achieved in 12 months with anti-TB treatment. Surprisingly, this patient's PPD test was anergic. Interestingly, the patient diagnosed with Brucellosis, who also had sacroiliitis, showed improvement with anti-TB treatment. This finding suggests that in cases of sacroiliitis that do not respond to appropriate treatment, other underlying factors may be involved. It is particularly important to consider the possibility of tuberculous sacroiliitis, especially in individuals residing in areas endemic for tuberculosis.

ETHICS COMMITTEE APPROVAL

This study was approved by Hatay Mustafa Kemal University Non-Invazive Clinical Research Ethics Committee (Decision no: 19, Date: 30.06.2022).

CONFLICT of INTEREST

The authors state no conflicts of interest.

AUTHORSHIP CONTRIBUTIONS

Concept and Design: TŞB, AKÇ

Analysis/Interpretation: TŞB, AKÇ

Data Collection or Processing: TŞB, AKÇ

Writing: TŞB, AKÇ

Review and Correction: TŞB, AKÇ

Final Approval: TŞB, AKÇ

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