

CAS CLINIQUE / CASE REPORT

SALMONELLA ENTERITIDIS BACTEREMIA WITH SEPTIC ARTHRITIS OF THE SACROILIAC JOINT IN A PATIENT WITH SYSTEMIC LUPUS ERYTHEMATOSUS **Case Report and Review of the Literature**

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El-Herte RI, Haidar RK, Uthman IW, Kanj SS. *Salmonella enteritidis* bacteremia with septic arthritis of the sacroiliac joint in a patient with systemic lupus erythematosus : Case report and review of the literature. J Med Liban 2011 ; 59 (4) : 235-237.

ABSTRACT : An 18-year-old female presented with a ten days history of high grade fever, chills and pain of the left sacroiliac joint. The patient has systemic lupus erythematosus (SLE) and is on chronic immunosuppressive therapy (steroids, antimalarial and antimetabolites). Imaging of the left sacroiliac joint revealed inflammation. Blood cultures and an aspirate of a small gluteal abscess that she developed later grew *Salmonella enteritidis* resistant to nalidixic acid. The patient was treated conservatively with eight weeks of IV ceftriaxone and is currently asymptomatic. First case of SLE with this complication to be reported from Lebanon and treated conservatively, this communication deserved publishing together with a literature review.

INTRODUCTION

Systemic lupus erythematosus (SLE) is a chronic inflammatory condition associated with systemic features and multiple organ involvement. Thus, these patients are prone to a variety of infections and complications [1]. Other than the disease flare, the differential diagnosis of a febrile patient with SLE and arthritis includes infection with different pathogens including *Salmonella* spp; *Salmonella* is endemic in our region [2-3]. The non-typhoid *Salmonella* (NTS) infections and their subsequent complications in these patients have been the most prevalent [4]. Septic arthritis is a known complication of NTS bacteremia mostly involving the hip, knees, and ankles [5]. The NTS bacteremia involving the sacroiliac joint is a rare complication in these patients [5]. Sacroiliac joint infection usually necessitates a combination of medical and surgical treatment [6-7]. The mainstay of treatment of septic arthritis is regular aspirations, surgical drainage or arthrotomy along with antimicrobial therapy [2, 4]. However, in certain situations where access to the joint is

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El-Herte RI, Haidar RK, Uthman IW, Kanj SS. Bactériémie à *Salmonella enteritidis* avec une arthrite de l'articulation sacro-iliaque chez un patient avec lupus érythémateux disséminé : A propos d'un cas. J Med Liban 2011 ; 59 (4) : 235-237.

RÉSUMÉ : Une jeune fille de 18 ans s'est présentée pour une fièvre de dix jours, frissons et douleur à la région sacro-iliaque gauche. La patiente est sous immunosuppression chronique pour lupus érythémateux disséminé (LED). La tomodensitométrie de l'articulation sacro-iliaque montre une inflammation non spécifique. La culture du sang et de l'abcès ultérieur formé à proximité de l'articulation sacro-iliaque gauche révèle *Salmonella enteritidis* résistant à l'acide nalidixique. La patiente a reçu un traitement conservateur par antibiotiques pour 8 semaines et est maintenant asymptomatique. Ce premier cas de LED avec cette complication au Liban et qui fut traité conservativement méritait d'être publié avec une discussion de la littérature.

difficult, more conservative options like intravenous antibiotic therapy or CT-guided drainage are chosen. With prompt treatment, clinical improvement is expected [2, 4]. We report a case of NTS sacroiliac joint infection in a patient with SLE managed medically without surgery.

CASE

An 18-year-old female presented in May 2010 to the emergency room with a ten days history of high grade fever with no focal complaints except for pain in the left hip area radiating to the corresponding lower extremity with inability to move the left hip because of pain. She did not report having any suspicious food intake and did not travel prior to her illness. The patient was diagnosed with SLE since the age of twelve and was maintained on prednisone 5 mg for more than one year, hydroxychloroquine 200 mg daily and methotrexate 10 mg weekly. Upon presentation to the emergency room, she was febrile (38.5 °C), tachycardic, hypotensive and the physical exam revealed an old systolic murmur grade 3/6 and tenderness of the left sacroiliac joint with decreased range of motion of the hip flexion. Laboratory tests showed anemia with a hemoglobin of 7.9 mg/dl; a WBC of 4000/ μ l with 89% neutrophils, and platelet count of 120.000/ μ l. Her creatinine was 1.7 mg/dl, with normal urine analysis and a normal chest X-ray. Blood cultures (BACT ALERT BioMerieux, France) were taken and the patient was started on moxifloxacin 400 mg per day, with a stress dose of hydrocortisone. Computed tomo-

graphy of the abdomen and pelvis showed splenomegaly with inflammation at the left sacroiliac joint consistent with inflammatory arthritis. Ten hours later the blood culture grew Gram negative rods identified as *Salmonella enteritidis* susceptible to ampicillin, cefotaxime, ceftazidime, ceftriaxone, ciprofloxacin and trimethoprim-sulfamethoxazole, and resistant to nalidixic acid. The patient was shifted to ceftriaxone 2 g every 12 hours which she received for seven days and was discharged home on per os cefixime 400 mg daily for 8 days. As soon as the patient discontinued the cefixime, the pain in the left gluteal area recurred. ESR was 54 mm/hr and CRP 7.3 mg/dl. Magnetic resonance imaging (MRI) showed a small abscess in the left gluteal region and a progression of the left sacroiliac joint inflammation. Drainage and culture of the gluteal abscess grew *Salmonella enteritidis*, with the same susceptibility pattern of the isolate recovered from the blood. Orthopedic surgery consult advised medical therapy with a prolonged course of antibiotics (ceftriaxone 2 g IV every 12 hours) which was given for 8 weeks, followed by trimethoprim-sulfamethoxazole double strength tablets twice daily for 4 weeks. Currently, after ten months the patient is asymptomatic walking free of pain with full range of motion.

DISCUSSION

NTS infections are acquired through the gastrointestinal tract and they usually cause gastroenteritis that is self-limited [5, 8]. Complications include gastroenteritis with bacteremia (5%), bacteremia alone without gastroenteritis, and bacteremia together with extraintestinal foci of infection (EFI) (7-12%). The latter includes: meningitis, diffuse colitis with crypt abscess, osteomyelitis, skin and soft tissue infection, endocarditis, urinary-tract infection, pneumonia, cholecystitis, and arthritis [5, 8, 10-11]. Several authors [3, 5, 12] have addressed the risk factors associated with NTS bacteremia with EFI. These factors [3, 5, 12] included: malignancy (23.6%-36.4%), diabetes mellitus (29.5%), immunosuppressive conditions, chronic kidney diseases, atherosclerosis and hypertension (27.9%-69.1%), HIV (18.6%-20%), connective tissue diseases (15.5%) among which 80% have SLE, age > 65 years, gastrectomy, chronic lung diseases, rheumatoid arthritis, amyloidosis, immunosuppressive therapy (steroids, cytotoxic chemotherapy), leukemia and lymphoma, and thalassemia [2, 5, 10, 12]. Our patient had a couple of these risk factors, namely: SLE, and chronic immune suppressive therapy. The EFI (30.9% to 39.5%) encompass a wide range of body sites [5, 10, 12]. In our case, the sacroiliac joint together with the gluteal area were the involved extraintestinal focal sites. An association between the immunosuppressive state and EFI with *Salmonella* spp. was noted by some authors [13-14]. Others, however, did not find a statistically significant difference in *Salmonella* infection among patients with and without immunosuppressive therapy (27.8% versus 36.8%) [5, 10]. In a review by Weinberger et al., the most commonly reported *Salmonella*

serotypes involved in bacteremia were: *S. enteritidis* (22.9%), *S. typhimurium* (20.1%), *S. hadar* (12.9%), *S. virchow* (12.2%) and *S. infantis* (6.9%) [11]. *S. enteritidis* and *S. typhimurium* were more prevalent with extreme of ages (< 2 years and > 60 years). In relation to children vs adults, *Salmonella* bacteremia was due to gastroenteritis in children and it was primary in adults. The most common serotype isolated was *S. enteritidis* in adults, and *S. virchow* in children [12]. *S. enteritidis* was also most commonly recovered from immunosuppressed (72%) vs immunocompetent patients (21.1%) ($p > 0.05$) [10]. With respect to SLE, the association with NTS infection has been studied. Hsu et al. [14] found a positive correlation between SLE and primary bacteremia with NTS. As reported by Chen et al. [5], SLE patients constitute 80% of the patients with connective tissue diseases who had NTS bacteremia. Moreover, in a hospital-based review of Gram-negative bacteremia, SLE was the most frequent underlying chronic disease [15]. Of these, the commonest bacterium was *Salmonella* (31.6%, 6 of 19) and SLE patients accounted for 20% of all non-typhoidal salmonellosis. When the joints were involved as EFI, infection of the hips was the most common (26 patients), followed by knees (11 patients), ankles (3 patients), sacroiliac joints (3 patients), wrists (1 patient), and elbows (1 patient) [4]. The predisposing risk factors for SLE patients to acquire NTS bacteremia and EFI include: high SLE disease activity index, high dose of immunosuppressive agents, lupus nephritis, anemia, and bone avascular necrosis. The most common *Salmonella* spp. serotypes isolated from these SLE patients were type B and D, and *S. enteritidis* was the most common pathogen causing septic arthritis in younger SLE patients [2, 4, 15-16]. The reverse association between recovery of *Salmonella* spp. and immune suppression was studied by Brown and Eykyn [17]. This was based on analysis of 82 cases of NTS bacteremia presenting to St. Thomas' Hospital between 1970 and 1999. The recovered salmonella were: *S. enteritidis* (48%), *S. typhimurium* (27%), and other serotypes (25%). Among these 82 patients, 59% had underlying immunosuppression, 80% had an extra-intestinal focus, and 80% of those with no focus had underlying immunosuppression. Thus, their advice was to search for immunosuppressive conditions in patients who present with NTS bacteremia without gastroenteritis [17]. The treatment of septic sacroiliitis consists of adequate antimicrobial coverage with or without surgical interventions. Antimicrobial treatment consists of four to six weeks of intravenous (IV) antibiotic therapy [7, 18-19]. Indications for surgical intervention include abscess formation, osteomyelitis, sequestrum of necrotic bone, and failure to respond to IV antibiotic therapy [19]. Most of the reports of septic sacroiliitis indicated the use of intravenous antibiotics with either abscess aspiration or debridement of the joint to get clinical improvement [7, 18-21]. Conservative management was reported in cases with intravenous antibiotics given for 6 and 8 weeks [6-7]. Our patient improved on IV antibiotics and aspiration of the soft tissue abscess without aspiration of the joint.

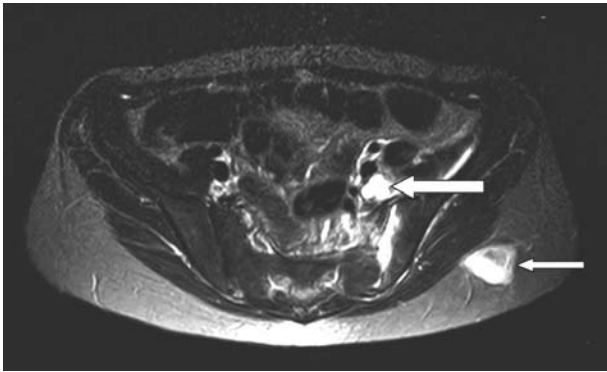


FIGURE 1. MRI showing abnormal high signal intensity on STIR images in the sacroiliac joint with enhancement postcontrast administration. A small abscess is seen just anterior and communicating with the sacroiliac joint on the left measuring 1.9 cm in diameter showing rim enhancement post contrast (upper arrow). Also an abscess is seen in the left gluteal area (lower arrow).

Contrast-enhanced MRI with subtraction technique may be useful for early detection of active sacroiliitis [22]. STIR sequences alone are sufficient to qualify and quantify an active sacroiliitis [23]. A contrast-enhanced MR sequence is beneficial to ensure maximum diagnostic confidence when patients with early sacroiliitis are examined. The MRI of sacroiliac joint in our patient (Figure 1) did not show any sequestra, necrosis or osteomyelitis, so the decision to treat conservatively was taken.

CONCLUSION

Septic arthritis in SLE patients should be considered in the differential diagnosis of a disease flare. When it occurs in immunocompromised patients, *Salmonella* should be suspected. Prompt identification of the causative pathogen should be sought in order to initiate early and appropriate antibiotic therapy.

REFERENCES

- Goldblatt F, Chambers S, Rahman A et al. Serious infections in British patients with systemic lupus erythematosus : hospitalisations and mortality. *Lupus* 2009 ; 18 : 682-9.
- Chen MJ, Tseng HM, Huang YL et al. Long-term outcome and short-term survival of patients with systemic lupus erythematosus after bacteraemia episodes : 6-yr follow-up. *Rheumatology* 2008 ; 47 : 1352-7.
- Bindal M, Krabak B. Acute bacterial sacroiliitis in an adult : A case report and review of the literature. *Arch Phys Med Rehabil* 2007 ; 88 : 1357-9.
- Chen JY, Luo SF, Wu YJJ, Wang CM, Ho HH. *Salmonella* septic arthritis in systemic lupus erythematosus and other systemic diseases. *Clin Rheumatol* 1998 ; 17 : 282-7.
- Chen PL, Chang CM, Wu CJ et al. Extraintestinal focal infections in adults with nontyphoid *Salmonella* bacteremia : predisposing factors and clinical outcome. *J Intern Med* 2007 ; 261 : 91-100.
- Roca B, Torres V. Pyomyositis of the iliacus muscle complicated with septic sacroiliitis. *Q J Med* 2008 ; 101 : 983-4.
- Yang SSY, Lee K. Unusual complication of intravenous Subutex abuse : two cases of septic sacroiliitis. *Singapore Med J* 2008 ; 49 (12) : 343-6.
- Gordon MA. *Salmonella* infections in immunocompromised adults. *J Infect* 2008 ; 56 : 413-22.
- Hohmann EL. Nontyphoidal salmonellosis. *Clin Infect Dis* 2001 ; 32 : 263-9.
- Dhanoa A. Non-typhoidal *Salmonella* bacteraemia : Epidemiology, clinical characteristics and its association with severe immunosuppression. *Ann Clin Microbiol Antimicrob* 2009 ; 8 : 15.
- Weinberger M, Andorn N, Agmon V, Cohen D, Shohat T, Pitlik S. Blood invasiveness of *Salmonella enterica* as a function of age and serotype. *Epidemiol Infect* 2004 ; 132 : 1023-8.
- Shimoni Z, Pitlik S, Leibovici L et al. Nontyphoid *Salmonella* bacteremia : Age-related differences in clinical presentation, bacteriology, and outcome. *Clin Infect Dis* 1999 ; 28 : 822-7.
- Yen YF, Wang FD, Chiou CC et al. Prognostic factors and clinical features of non-typhoid *Salmonella* bacteremia in adults. *J Chin Med Assoc* 2009 ; 72 : 408-13.
- Hsu RB, Tsay YG, Chen R, Chu SH. Risk factors for primary bacteremia and endovascular infection in patients without acquired immunodeficiency syndrome who have nontyphoid salmonellosis. *Clin Infect Dis* 2003 ; 36 : 829-34.
- Lim E, Koh WH, Loh SF, Lam MS, Howe HS. Nontyphoidal salmonellosis in patients with systemic lupus erythematosus : A study of fifty patients and a review of the literature. *Lupus* 2001 ; 10 : 87-92.
- Huang JL, Hun JJg, Wu KC, Lee WI, Chan CK, Ou LS. Septic arthritis in patients with systemic lupus erythematosus : *Salmonella* and non-*Salmonella* infections compared. *J Sem Arthritis* 2006 ; 4 : 61-6.
- Brown M, Eykyn SJ. Non-typhoidal *Salmonella* bacteraemia without gastroenteritis : a marker of underlying immunosuppression. Review of cases at St. Thomas' Hospital 1970-1999. *J Infect* 2000 ; 41 : 256-9.
- Zimmermann B III, Mikolich DJ, Lally VE. Septic sacroiliitis. *Sem Arthritis Rheumat* 1996 ; 26 : 592-604.
- Almoujahed MO, Khatib R, Baran J. Pregnancy-associated pyogenic sacroiliitis : case report and review. *Infect Dis Obstet Gynecol* 2003 ; 11 : 53-7.
- Moros ML, Rodrigo C, Villacampa A, Ruiz J, Lapresta C. Septic shock in pregnancy due to pyogenic sacroiliitis : a case report. *J Med Case Reports* 2009 ; 3 : 6505.
- Chan DS, Saklani A, Shah PR, Haray PN. Laparoscopic drainage of retroperitoneal abscess secondary to pyogenic sacroiliitis. *Ann R Coll Surg Engl* 2010 ; 92 : 32-4.
- Oktay A, Gokhan G, Bulent B, Gokhan O, Zeynep Y. Evaluation of sacroiliitis : contrast-enhanced MRI with subtraction technique. *Skeletal Radiol* 2009 ; 38 : 983-8.
- Althoff CE, Feist E, Burova E et al. Magnetic resonance imaging of active sacroiliitis : Do we really need gadolinium ? *European Journal of Radiology* 2009 ; 71 : 232-6.