Case Report
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Enterobacter cloacae Sacroiliitis with Acute Respiratory Distress Syndrome in an Adolescent

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Enterobacter cloacae has emerged as an important nosocomial pathogen, but is rarely a cause of sacroiliitis. Herein, we present the first reported case of *Enterobacter cloacae* sacroiliitis associated with sepsis and acute respiratory distress syndrome (ARDS). A previously healthy 14-year-old boy presented with low-grade fever and pain in the left side of the hip that was aggravated by walking. Pelvic computed tomography (CT) showed normal findings, and the patient received supportive care for transient synovitis with no antibiotics. However, there was no clinical improvement. On the third day of hospitalization, magnetic resonance imaging of the hip revealed findings compatible with sacroiliitis, for which vancomycin and ceftriaxone were administered. The patient suddenly developed high fever with dyspnea. Chest radiography and CT findings and a PaO_2/FiO_2 ratio <200 mmHg were suggestive of ARDS; the patient subsequently received ventilatory support and low-dose methylprednisolone infusions. Within one week, defervescence occurred, and the patient was able to breathe on his own. Following the timely recognition of, and therapeutic challenge to, ARDS, and after 6 weeks of parenteral antimicrobial therapy, the patient was discharged in good health with no complications.

Key Words: *Enterobacter cloacae*; Sepsis; Sacroiliitis; Acute respiratory distress syndrome; Adolescent

Introduction

Staphylococcus aureus is the most common causative organism of sacroiliitis [1]. Only a few gram-negative bacteria have been found to be the causative pathogens in sacroiliitis [2-4]. Sacroiliitis manifesting with acute respiratory distress syndrome (ARDS) has rarely been reported: one case caused by *S. aureus* and the other by group G *Streptococcus* [5, 6]. *Enterobacter cloacae* has been known to cause various nosocomial infections with high rates of morbidity and mortality, but cases of sacroiliitis caused either by *E. cloacae* alone, or associated with sepsis and ARDS, have not previously been reported. The following is a case of *E. cloacae* sacroiliitis associated with sepsis and ARDS in a previously healthy boy who was successfully treated with antibiotics, respiratory support, and low-dose corticosteroid infusions.

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Case Report

A 14-year-old boy was admitted to the Department of Orthopedic Surgery with a 1-day history of fever and pain in the left side of the hip that was aggravated by walking. The patient denied any injuries, medical problems, intake of any medications, previous hospitalizations, or recent tours. His family history was unremarkable with respect to musculoskeletal disorders. Although his temperature was 38°C, he did not appear ill. Other observations were as follows: blood pressure, 110/70 mmHg; heart rate, 88/min; and respiratory rate, 22/min. He experienced tenderness over the lateral aspect of the left side of the hip, which was accentuated by flexion and external rotation. Initially, laboratory studies showed a hemoglobin level of 14.6 g/dL, a white blood cell (WBC) count of 6,400/mm^3 with 79% neutrophils, platelet count of 176,000/mm^3, erythrocyte sedimentation rate (ESR) of 12 mm/h, and C-reactive protein (CRP) level of 3.1 mg/dL (normal range, <0.3 mg/dL). Hip radiography and pelvic computed tomography (CT) revealed no abnormal findings. Initially, transient synovitis was considered, and antibiotics were therefore not prescribed. On the third hospital day, the body temperature had risen to 38.9°C, and physical examination revealed pain at the sacroiliac joint on the FABERE (flexion–abduction–external rotation–extension) test. A hip magnetic resonance imaging (MRI) scan showed synovitis and capsulitis of the left sacroiliac joint with extra-articular extension to the left iliacus muscle and iliac vessels, suggesting sacroiliitis (Fig. 1A). Empiric administration of vancomycin (4 g/day) and ceftriaxone (4 g/day) was initiated to treat the sacroiliitis. The patient was transferred to the Department of Pediatrics where his condition deteriorated rapidly, with his fever reaching 40.4°C and respiratory rate being >40/min with difficulty breathing. An arterial blood gas analysis (ABGA) showed the following results: pH, 7.399; pCO_2, 32.7 mmHg; PO_2, 52.7 mmHg; and O_2 Sat, 88%. Dyspnea and ABGA findings were initially improved with oxygen supply via a nasal cannula. The respiratory status, however, worsened the following day, as the ABGA showed a PaO_2/FiO_2 of 137 mmHg, chest radiographs began to show bilateral lower lobe infiltrates (Fig. 2 A), and chest CT (Fig. 2 B) showed bilateral and symmetric compartmental consolidation in gravity-dependent areas of the lung compatible with ARDS, which was not observed over the thorax on the abdominal CT image obtained on the first hospital day. The patient was moved to the intensive care unit (ICU) for synchronized intermittent mandatory ventilation with pressure support; the initial settings used were a tidal volume of 360 mL (6 mL/kg), FiO_2 of 40%, and positive end-expiratory pressure (PEEP) of 12 cmH_2O. WBC and platelet counts decreased to 2,400/mm^3 with 79% neutrophils and 58,000/mm^3, respectively, and ESR and CRP were increased to 40 mm/h and 13.8 mg/dL, respectively. Blood cultures, which were taken on the second hospital day, yielded *E. cloacae* which is susceptible to amikacin, aztreonam, ceftriaxone, ceftazidime, cefotaxime, ciprofloxacin, gentamicin, imipenem, piperacillin/tazobactam; intermittently resistant to cefuroxime; and resistant to amoxicillin/clavulanic acid, ampicillin, ampicillin/sulbactam, cephalothin, cefoxitin, and cefazolin. The antibiotic regimen was changed to ceftriaxone (4 g/day) and amikacin (1.5 g/day). Because of the rapid progression of ARDS, methylprednisolone (1 mg/kg) administration was initiated. By the fifth day, defervescence occurred and the need for ventilatory support was reduced. On the seventh day, the patient was breathing room air and the pain in the left side of the hip lessened dramatically; he was subsequently discharged from the ICU. A follow-up chest radiograph demonstrated marked improve-

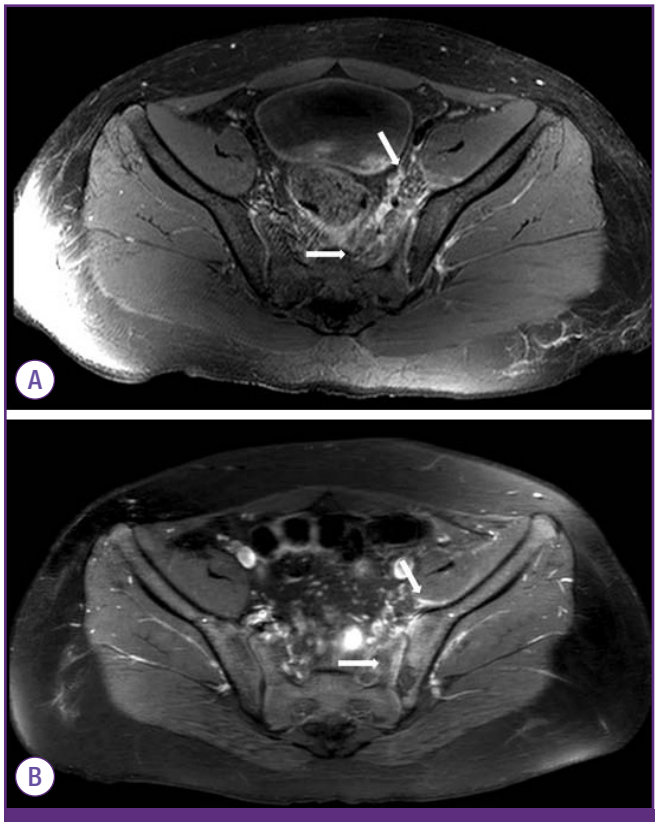

Figure 1. Fat-suppressed axial T1-weighted magnetic resonance images of sacroiliac joints showing synovitis and capsulitis of the left sacroiliac joint with extra-articular extension to the left iliacus muscle and iliac vessels on the third hospital day (A) and improvement in the extra-articular soft tissue inflammation but aggravated subchondral inflammation on the 13th hospital day (B) (arrow).

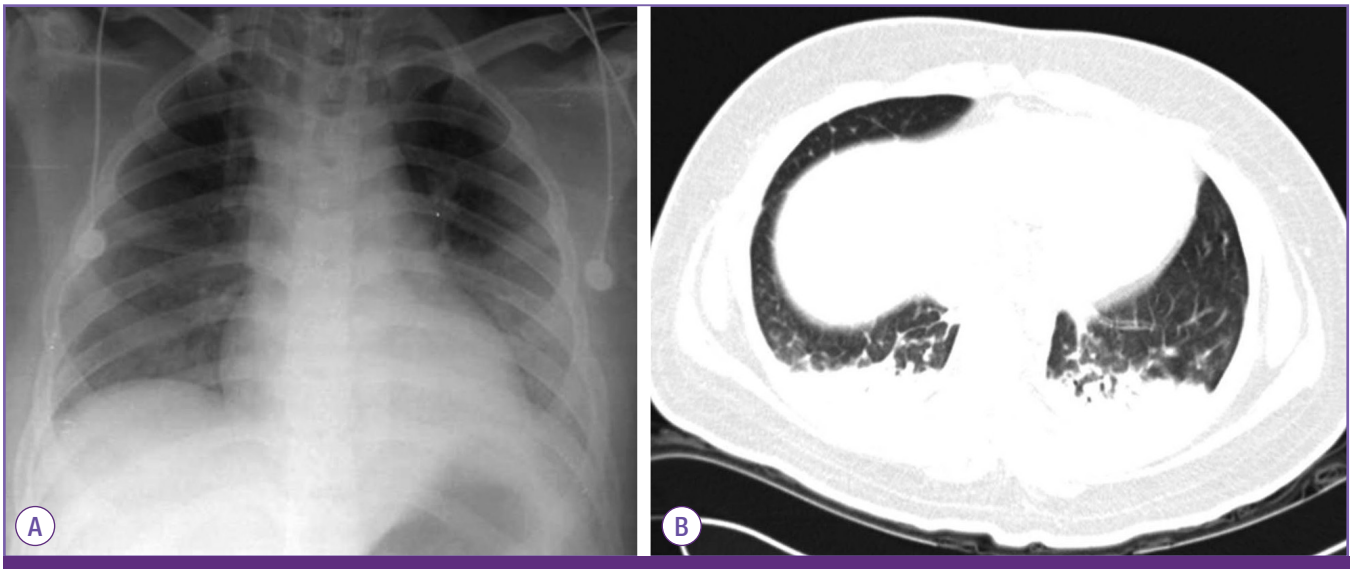

Figure 2. Chest radiograph on the fourth hospital day showing bilateral lower lobe infiltrates (A). Chest computed tomography images on the third hospital day shows bilateral and symmetric compartmental consolidation in gravity-dependent areas of the lung compatible with acute respiratory distress syndrome (B).

ment. Parenteral antimicrobial therapy was maintained for 6 weeks until CRP and ESR normalized, and low doses of methylprednisolone were administered and tapered over 19 days. A repeat hip MRI scan on the thirteenth hospital day showed improvement in extra-articular soft tissue inflammation but with aggravated subchondral inflammation (Fig. 1B). Repeat blood culture on the fourth hospital day grew no bacteria, and the patient was discharged on day 46 in good health without any complications. Serum immunoglobulin and complement levels were normal: IgG 890 mg/dL, IgA 83.3 mg/dL, IgM 86.3 mg/dL, C3 144.0 mg/dL, and C4 22.4 mg/dL, and no further tests for an immunologic disorder were pursued since the patient had shown marked improvement under treatment.

Discussion

Pyogenic sacroiliitis is relatively rare, representing only 1.5% of all cases of pyogenic arthritis in children [1]. Its diagnosis may be delayed, or misdiagnosed as synovitis, sciatica, acute abdomen, or septic arthritis of the hip, due to poor localized initial symptoms and inadequate physical examination [3, 7]. A positive FABERE test for the sacroiliac joints strongly suggests the possibility of sacroiliac disease [8]. Most sacroiliac joint infections appear to occur via hematogenous spread, although the frequency of obvious preceding infections varies. In a review of 163 cases of sacroiliac joint infection from 1878 to 1990, it was noted that 41% of the patients demonstrated no associated factors; 75% had an acute presentation such as high fever, evident infection, and severe continuous pain exacerbated by weight bearing or movement of the sacroiliac joint; and 25% had a subacute presentation with or without a low-grade fever and less intense pain [9]. Radiographs of the sacroiliac joint are usually normal during the early course of the disease, and MRI is more sensitive than CT for detecting infectious sacroiliitis because of its superior evaluation of cartilage integrity and detection of osseous erosions [10]. This patient, who had an acute presentation with fever, hip pain, and a positive FABERE test, was diagnosed with sacroiliitis after a hip MRI, not by hip radiographs or pelvic CT taken earlier.

Although *S. aureus* is the most common causative organism of sacroiliitis,-gram negative bacteria including *Haemophilus influenza, Citrobacter freundii, Salmonella* spp, and *Brucella melitensis* have occasionally been the cause of sacroiliitis [2-4, 11]. Furthermore, only two cases of sacroiliitis associated with ARDS have been reported: an adolescent with group G *Streptococcus* sepsis [6] and an adult with *S. aureus* septicemia [5]. Even though *E. cloacae* is an increasingly common nosocomial pathogen in children, especially for pneumonia and serious infections such as sepsis in neonatal intensive-care units, surgical wards, and burn units [12], this patient represents the first reported case of *E. cloacae* sacroiliitis with sepsis and ARDS. Under the new Berlin definition, the patient had acute onset of moderate ARDS with bilateral opacities detected on chest CT and chest radiography as well as a PaO_2/FiO_2 of 137 mmHg with a PEEP of 12 cmH_2O [13]. Coordinated

management by incorporating a combination of antimicrobial therapy (ceftriaxone and amikacin), protective ventilation support, and the early use of low-dose methylprednisolone appears to have shortened the need for ventilatory support to only 4 days and hastened the recovery, when the risk of mortality and the median duration of mechanical ventilation in patients with moderate ARDS are 32% (95% confidence interval, 29–34%) and 7 days (interquartile range, 4–14 days) [13-16], respectively.

In conclusion, it should be kept in mind that sacroiliitis can, though rarely, manifest with sepsis and ARDS in previously healthy children. When it occurs, early recognition and optimal choice of antibiotics along with timely aggressive supportive care with mechanical ventilation and methylprednisolone may cure sacroiliitis and prevent long-term debilitating effects of rapidly progressing ARDS due to *E. cloacae* sepsis.

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