

Pseudo-septic hip arthritis as the presenting symptom of ankylosing spondylitis: a case series and review of the literature

T. Mathur, A.M. Manadan,
B. Hota, J.A. Block

Rush University Medical Center and John H. Stroger, Jr. Hospital of Cook County, Chicago, Illinois, USA.

Tanisha Mathur, Resident Physician
Augustine M. Manadan, Associate Professor
of Medicine

Bala Hota, Assistant Professor of Medicine
Joel A. Block, Professor of Medicine

Please address correspondence
and reprint requests to:

Augustine M. Manadan, MD,
Rush University Medical Center,
1725 W. Harrison, Suite 1017,
Chicago, IL 60612, USA.

E-mail: amanadan@rush.edu

Received on January 18, 2010; accepted
in revised form on March 22, 2010.

© Copyright CLINICAL AND
EXPERIMENTAL RHEUMATOLOGY 2010.

Key words: Ankylosing spondylitis,
pseudoseptic hip arthritis

ABSTRACT

Introduction. *Ankylosing spondylitis (AS) typically presents with inflammatory back pain and stiffness, but severe hip involvement may also be present. In this study, we evaluated the frequency of severe hip arthritis mimicking septic arthritis as initial presenting symptom of AS*

Methods. *Utilising billing records, we retrospectively studied all AS patients seen from the years 2006 to 2009 at our institution. The primary endpoint was severe hip arthritis mimicking septic arthritis as the initial presenting symptom of AS.*

Results. *A total of 121 AS patients were identified from billing records, of whom 3 had severe hip arthritis mimicking septic arthritis as the initial presenting symptom of ankylosing spondylitis.*

Conclusions. *Our study highlights the importance of including AS in the differential diagnosis of severe acute inflammatory hip arthritis in young adults, even when the onset appears to be abrupt.*

Introduction

Ankylosing spondylitis (AS) is a seronegative spondyloarthropathy characterised by axial arthritis, ocular inflammation, enthesitis, and peripheral oligoarthritis (1, 2). AS has a predilection for young adults, a slight male predominance, and usually presents with inflammatory back pain (1-3). Although the hips are frequently involved during the course of AS (4), initial presentation with severe hip arthritis is unusual, and abrupt-onset hip arthritis mimicking septic arthritis in AS has not been widely appreciated. Here, we report a series of patients with AS who presented initially as pseudo-septic hip arthritis.

Methods

After obtaining institutional review board approval, the charts of all patients who were diagnosed with AS in the adult rheumatology clinics at John H Stroger, Jr. Hospital of Cook County (Chicago, IL) from the years 2006 to August 2009 were retrospectively evaluated. Patients were identified electronically by ICD-9 codes 720.0 from billing records. The primary endpoint was pseudoseptic hip

arthritis as the initial presenting symptom of AS. "Pseudoseptic" was defined as a clinical presentation mimicking bacterial septic arthritis. The diagnosis of AS was confirmed using the modified New York criteria (5), and then all records were reviewed for demographic information, presenting complaints, and radiologic studies. Additionally, we conducted a literature search in PubMed combing the search terms "pseudoseptic, pseudosepsis, ankylosing spondylitis, seronegative spondyloarthropathy, and hip pain."

Results

A total of 121 patients were identified from billing records. After detailed review of medical records, 83 patients met the modified New York criteria (5) for definite AS and 27 met criteria for probable AS. Of these 110 patients, eleven presented with severe hip arthritis as the primary reason for referral to rheumatology. Three of the eleven had severe hip arthritis mimicking septic arthritis at the time of referral. Details of those three cases are as follows:

Case 1

A 22-year-old Hispanic male immigrant presented to the emergency department with acute worsening of chronic right hip pain radiating down the right leg. He had experienced hip pain for 6 years but over last several months required crutches for ambulation. He was found to have a positive purified protein derivative (PPD). He reported subjective fevers. Examination revealed point tenderness over sacroiliac joints, the lumbar spine and the sacrum. Right hip flexion was limited. Laboratory evaluation revealed elevated erythrocyte sedimentation rate (ESR) of 93 mm/hr. and a high C-reactive protein (CRP) of 5.91 mg/dl. Computerised axial tomography (CT) of the hip showed severe uniform narrowing of the right hip with erosive changes in the acetabular roof and severe narrowing with erosion and sclerosis of the sacroiliac joints. A magnetic resonance imaging (MRI) scan of the hip showed narrowing of the right hip with thickening of the synovium, destructive lesions and abnormal marrow replacement of the acetabulum and

Competing interests: none declared.

femoral head. Inflammatory changes were also seen in the femoral neck, lesser trochanter and bilateral trochanteric bursae. The radiologist reported, "Bilateral hips with infectious arthritis and osteomyelitis, greater on the right. Correlate clinically for possible tuberculous arthritis/osteomyelitis." Arthrocentesis of the right hip was performed. No cell count was performed; Gram's stain and Acid-fast stain were negative, and culture negative. The patient was subsequently referred to rheumatology clinic and diagnosed with AS. Etanercept 50 mg subcutaneously (SC) per week was initiated, and the patient improved dramatically; after 12 weeks, he was ambulating without crutches.

Case 2

A 21-year-old Hispanic female immigrant presented with a 2-week history of pain and weakness in both legs affecting the ankle, knee and hip joints. She also described swelling of the left second and third toes associated with subjective fevers and chills. Laboratory evaluation revealed ESR of 118 mm/hr., CRP of 23 mg/dl, and positive human leukocyte antigen (HLA) B27. MRI scan showed bilateral narrowing of hip joint space associated with thickening and contrast enhancement of synovium. Destructive lesions with abnormal marrow replacement of the left acetabular roof and femoral head and neck were also seen. The radiologist's interpretation was "suggestive of septic arthritis of bilateral hips and osteomyelitis of femoral head and acetabulum." Arthrocentesis of the hip revealed 945 red blood cells/mm³ (RBC) and 5,251 white blood cells/mm³ (WBC) with 80% polymorphs. Culture of the synovial aspirate was negative. The patient received intravenous antibiotics for 6 weeks for presumed septic arthritis but returned soon after to the hospital with recurrent left hip and left knee pain. On repeat admission, the rheumatology service diagnosed undifferentiated spondyloarthritis based on hip arthritis, heel pain, and inflammatory oligoarthritis, and initiated therapy with methotrexate and oral prednisone. After tapering of the prednisone, the severe bilateral hip pain recurred. A repeat hip

arthrocentesis had 130,000 RBCs/mm³ and 8140 WBCs/mm³ with 86% polymorphs. The patient was lost to follow-up for one year, after which she presented with worsening of hip and knee pain and inability to ambulate. Repeat CT scan showed symmetrical sacroiliitis, moderate bilateral narrowing and erosions of both hip joints which were most consistent with a diagnosis of AS. Etanercept 50 mg SC weekly was begun, and the patient had virtually complete resolution of pain and regained the ability to ambulate.

Case 3

A 23-year-old Hispanic male immigrant presented to another hospital with a 3-year history of left hip pain, progressing recently to lower back pain. MRI scan showed left hip effusion and marrow edema of the ileum and ischium. PPD was positive. Synovial biopsy was performed to exclude tuberculous septic arthritis. Cultures were negative and no granulomas were found. The patient was referred to rheumatology at our institution. Physical examination revealed severe limitation of left hip motion and an abnormal Schoeber's test (10.5/10cm). CT scan showed bilateral sacroiliitis. The patient was treated with etanercept 50 mg SC weekly for AS and isoniazid for latent tuberculosis, with resolution of back and hip symptoms.

Results of the literature review

The results of the PUBMED literature review yielded no relevant articles when the search terms "pseudoseptic, pseudosepsis were combined with ankylosing spondylitis or seronegative spondyloarthropathy."

Discussion

Hip involvement in AS is common and often disabling, whereas involvement of the other root joint, the shoulder, is less frequent and less severe. Corrected for disease duration and age at onset, patients with severe radiological hip involvement are more prone to have severe axial disease (OR 5.5 95% CI 3.1–9.5, $p < 0.001$) (6). Current literature suggests that severe involvement of hip in AS patients tends to occur at a younger age and is correlated with

longer disease duration. Histopathologically, hip involvement in AS seems to be largely based on inflammation of the subchondral bone marrow (7). However, in contrast to the classical changes of AS in the spine, rheumatic inflammation in the hip does not lead to formation of new bone, but results in an erosive disease, which will often destroy the joint (7).

Despite the fact that hip involvement is common in AS, there is little literature concerning the severity of hip involvement at presentation, and we found no prior reports of pseudoseptic hips in this patient population. We found that 10% (11 of 110) of patients with newly diagnosed AS at our institution presented with severe hip involvement, and three of those presented with symptoms of acute-onset hip pain with clinical and laboratory features that mimicked bacterial arthritis, so-called "pseudoseptic arthritis" (8). Patients with pseudoseptic arthritis are typically treated initially as if they have an infectious etiology, with a complete search for the source and portal of entry of the infection, and broad-spectrum antibiotic therapy is typically initiated if the synovial fluid leukocyte count is high with neutrophil predominance. In the setting of sterile cultures and rapid clinical improvement, pseudoseptic arthritis as a likely explanation, should be considered. Failure to do so can result in significant delays in reaching the appropriate diagnosis and initiating treatment, as seen in our three patients.

Although there have been previous case reports in patients with rheumatoid arthritis (8), our study is the first to report the frequency (3/121) of pseudoseptic arthritis as the initial presentation of AS. We propose that AS, in addition to septic arthritis, should be considered in the differential diagnosis of severe acute inflammatory hip arthritis in young adults.

References

1. KLIPPEL JH, CROFFORD LJ, STONE JH *et al.*: Primer on the rheumatic diseases. Atlanta, Georgia, Arthritis Foundation 2002.
2. KHAN MA: Update on spondyloarthropathies. *Ann Int Med* 2002; 136: 886-906.
3. VAN DER HORST-BRUIJNSMA IE, LEMS WF, DIJKMANS BAC: "A systematic comparison

- of rheumatoid arthritis and ankylosing spondylitis." *Clin Exp Rheumatol* 2009; 27 (Suppl. 55): S43-49.
4. CALIN A, ELSWOOD J: The relationship between pelvic, spinal and hip involvement in ankylosing spondylitis-one disease process or several? *Br J Rheumatol* 1988; 27: 393-5.
 5. LINDEN VD: Evaluation of diagnostic criteria for ankylosing spondylitis. A proposal for modification of the New York criteria. *Arthritis Rheum* 1984; 27: 361-8.
 6. CRUYSSSEN B, MUNOZ-GOMARIZ E, FONT P *et al.*: Hip involvement in ankylosing spondylitis: epidemiology and risk factors associated with hip replacement surgery. *Rheumatology* 2009; 48: 1-6.
 7. APPEL H, KUHNE M, SPIEKERMANN S *et al.*: Immunohistochemical analysis of hip arthritis in ankylosing spondylitis: evaluation of the bone-cartilage interface and subchondral bone marrow. *Arthritis Rheum* 2006; 54: 1805-13.
 8. CALL RS, WARD JR, SAMUELSON CO JR: 'Pseudo-septic' arthritis in patients with rheumatoid arthritis. *West J Med* 1985; 143: 471-3.