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Tuberculous sacroiliitis. A case report and review of the literature

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Abstract *Background:* Infections of the musculoskeletal system are tuberculous in nature in 1–5% of cases. The sacroiliac joint is involved in 3–9.7%. We describe the case of a 32-year-old man with tuberculous sacroiliitis presented as a growing mass on the lateral aspect of his right proximal and mid-thigh. Open biopsy, histology, cultures and PCR established the diagnosis of tuberculosis. *Results:* After surgical drainage of the abscess, the patient was administered a triple antibiotic regimen for 12 months. Seven years postoperatively, the patient is dis-

ease-free with no functional limitation. This case report highlights the importance of continued awareness for early detection and treatment of a tuberculous sacroiliac joint infection.

Keywords Skeletal tuberculosis · Abscess · Tuberculous sacroiliitis · Tuberculosis of the greater trochanter

Introduction

Tuberculosis affects worldwide one-third of the population, being the most common infectious cause of death [1]. In contrast, a decreased incidence in the United States is reported [10] after an almost 2 decade interval of increased rates [9]. In urban areas, the incidence of the disease is comparable to that of developing countries, as a result of the high-risk population segregation, and the emergence of drug-resisting bacilli due to the injudicious use of antibiotics and the lack of patient compliance [39].

In the 1950s, because of the optimism succeeding the development of antibiotics, tuberculosis was often overlooked as a cause of musculoskeletal infections [40]. The lack of suspicion and the scant experience of the medical community accounts for the average delay of 17 months between the initial symptoms and the definite diagnosis of skeletal tuberculosis [39, 40].

Infections of the musculoskeletal system are tuberculous in nature in 1–5% of cases [8, 14]. The sacroiliac joint is involved in 3–9.7% [2, 6, 14, 20, 30]. Of note is the fact that the reported incidence of sacroiliac joint tuberculosis is negligible in developing countries [26].

Sacroiliac joint infections represent a diagnostic dilemma for their vague and non-specific clinical presentation [17]. Herein, the authors report a patient with sacroiliac joint tuberculous infection that was undiagnosed for several years, until the infection spread to the greater trochanter and an ipsilateral thigh abscess developed.

Case report

In July 1997, a 32-year-old man, a schoolteacher in a rural area, was admitted to the authors' institution

complaining of right buttock pain and an enlarging mass over the lateral aspect of his right hip and mid-thigh.

His pain originated in 1992 after a fall. At that time, plain radiographs of the pelvis showed no fractures; however, there were extensive erosions at the right sacroiliac joint and faint calcifications at the ipsilateral ischial tuberosity (Fig. 1). Tc^{99m}-bone scintigraphy demonstrated increased radioisotope uptake at the right sacroiliac joint and the ipsilateral greater trochanter (Fig. 2). His local physician attributed the sacroiliac joint lesion to a previous *Brucella mellitensis* infection and the increased radioisotope uptake at the



Fig. 1 Anteroposterior radiograph of the pelvis in a 27-year-old man, showing erosions of the right sacroiliac joint and demarcation of the cortex of the right ischial tuberosity



Fig. 2 Bone scintigraphy with ^{99m}Tc MDP exhibiting increased radioisotope uptake at the right sacroiliac joint, the right trochanteric area and ischial tuberosity

region of the greater trochanter to his fall. The patient refused any further investigation because his pain was adequately relieved with non-steroid anti-inflammatory drugs.

In December 1996, the patient's buttock pain had worsened and an enlarging thigh mass emerged. The pain radiated to the ipsilateral calf, especially during climbing stairs and intense walking. The patient had sporadic systemic episodes of low-grade fever and night sweats. Past medical history included positive agglutination tests for brucellosis, for which the patient had received oral tetracyclines. Interestingly, before the definite diagnosis of brucellosis, a 3-week course of intramuscular streptomycin had been administered with improvement of his symptoms. Family history was negative for tuberculosis.

At his admission, the patient was walking with an antalgic gait. Clinical examination revealed a painless mass over the lateral aspect of his right hip and mid-thigh and moderate swelling at the inner thigh and the ipsilateral perineal area. The right sacroiliac joint was tender in deep palpation and stretching maneuvers, including lateral pelvic compression test, Patrick and Gaenslen's tests. Right hip joint motion was painful at the extremity. Right straight leg raising test was positive at 50°. There were no motor or sensitivity neurologic deficits in the lower extremities.

Plain anteroposterior radiograph of the pelvis showed a sacroiliac joint lesion and significant erosions of the right greater trochanter (Fig. 3). Computed tomography (CT) of the pelvis also showed the right sacroiliac joint erosions, in addition to scattered calcifications and extensive destruction of the ipsilateral greater trochanter and ischium (Fig. 4). Magnetic



Fig. 3 Preoperative anteroposterior radiograph of the pelvis showing destructive lesion of the right trochanter and right ischial tuberosity

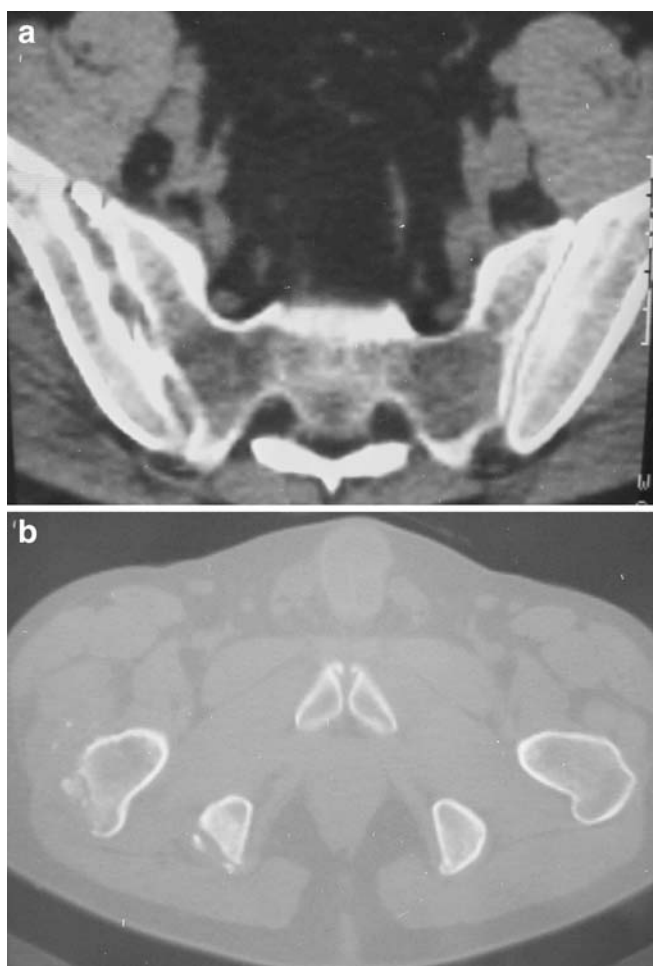


Fig. 4 **a** Axial computer tomography image showing erosions of the articular surface of the right sacroiliac joint. **b** Axial CT-image depicting destruction at the right trochanter and ischial tuberosity. Soft tissue calcifications are delineated in both sites

resonance imaging (MRI) demonstrated a large soft tissue mass at the right mid-thigh, with signal abnormalities suggesting an abscess (Fig. 5). Chest radiographs, and CT and MRI of the lumbar spine were negative.

Erythrocyte sedimentation rate (ESR) was 5 mm/h and C-reactive protein (CRP) was 0.9 mg/dl (normal value, <0.5 mg/dl). There was a mild hypochromic and microcytic anemia; hematocrit was 39.5%. Slide agglutination tests with *Brucella* antiserum were negative. Tuberculin skin test was positive (10 mm); however, the patient had a history of BCG vaccination 10 years previously.

Open biopsy was performed through a lateral approach over the right greater trochanter and extensive debridement was performed. The gross specimen was soft, friable, whitish-gray clumped cheese-like material. Direct smear and stain of the material showed acid-fast



Fig. 5 **a** Coronal T-2 weighted magnetic resonance image of the pelvis showing a soft tissue mass at the right hip and proximal thigh, associated with destruction and significant edema of the right trochanter. **b** Axial T-2 weighted magnetic resonance image showing a soft tissue mass of the right proximal thigh

bacilli. Histological examination was consistent with a granulomatous infection. Polymerase chain reaction (PCR) showed amplification of the *M. tuberculosis* genome. Cultures in Löwestein–Jensen culture medium isolated *M. tuberculosis*, and the diagnosis of tuberculosis was confirmed.

Postoperatively, the patient was administered a triple anti-tuberculous chemotherapy regimen including isoniazid (5 mg/kg body weight), rifampicin (10 mg/kg body weight), and pyrazinamide (35 mg/kg body weight) for 12 months. Within this year, clinical symptoms resolved completely, and MRI showed complete regression of the abscess. Seven years later, the patient is pain-free with no functional limitations. Plain radiograph of the pelvis demonstrates spontaneous ankylosis of the sacroiliac joint (Fig. 6).



Fig. 6 Postoperative anteroposterior radiograph of the pelvis 5 years after the treatment. The sacroiliac joint is spontaneously ankylosed; there are residual changes of the right trochanter and ischial tuberosity

Discussion

The diagnosis of sacroiliac joint infection is often delayed. In the current patient, the short-term administration of streptomycin masked the clinical presentation of tuberculous sacroiliac joint infection, which has been spread in continuity to the ipsilateral greater trochanter. A delayed diagnosis may also be attributed to the inaccessibility of the sacroiliac joint while the patient is examined in the supine position [17], as well as to the physicians' failure to perform the sacroiliac joint pain provocation maneuvers [12].

Frequently, mycobacterial infections of the sacroiliac joint may present as a psoas abscess, and may not be diagnosed until spontaneous drainage occurs in the groin [16, 21, 31, 32, 34]. The enlarging thigh abscess was the presenting symptom in the present case, ensuing from pus released after the rupture of the anterior sacroiliac joint capsule.

Buttock pain is invariably present in tuberculous sacroiliitis [11, 15–17, 21, 31, 32, 34, 38]. The sacroiliac pain can be referred to the groin, posterior thigh, and occasionally below the knee, mimicking pain originating from the lumbar spine, the hip and the lower abdominal quadrant [4, 5, 12, 15, 16, 32, 37]. There may be signs of femoral or sciatic nerve root irritation if the distended anterior joint capsule comes in contact with the lumbosacral plexus [3]. In a series, Richter et al. [34] reported that 92% of patients were misdiagnosed and were initially treated for lumbosacral radiculopathy, or underwent useless lumbar spine surgery.

Pain is usually reproduced or aggravated when the sacroiliac joint is mechanically strained during physical examination [23], weight bearing, bending, sitting, or riding [7, 11, 12, 21, 25, 32, 34], and is frequently alleviated by standing or walking. Rarely are these associated neurologic symptoms of weakness, paresthesias, or dysthesias [3, 11]. Tenderness on rectal examination is possible [17, 24]. The trochanteric lesion is usually not associated with local pain [7, 22, 25].

Routine laboratory tests such as elevated ESR and CRP are not significant in the diagnosis of tuberculosis, but are considered useful in the assessment of the response to anti-tuberculous therapy [20]. The ESR of the current patient was significantly lower than normal, suggesting a non-pyogenic infection.

Plain radiographs may not show any abnormality in the early stage of sacroiliac tuberculosis. Haziness or loss of joint line may be an early radiographic finding. Bone scan is helpful for early recognition of the condition [35]. However, CT or MRI are more helpful for early detection of sacroiliac tuberculosis [20, 21]. CT scan shows clearly the extent of joint destruction and MRI delineates the abscess in the soft tissues [20, 21, 24, 32, 38]. MRI may also contribute to the differential diagnosis from a soft tissue tumor or pyogenic arthritis [19].

Tuberculous sacroiliitis should be differentiated from degenerative and post-traumatic arthritis; pyogenic infection of the sacroiliac joint; inflammatory diseases such as sero-negative spondyloarthropathies, ankylosing spondylitis, psoriatic arthritis, Reiter's and Behçet's syndromes and inflammatory bowel diseases; connective tissue disorders such as rheumatoid arthritis and systemic lupus erythematosus; familial Mediterranean fever with multisystem involvement including arthritis; osteitis condensans ilii; metabolic conditions such as gout pseudogout and hyperparathyroidism; tumors and tumorlike conditions, and pigmented villonodular synovitis [3, 13, 20, 24, 29, 33].

Pyogenic infection of the sacroiliac joint is usually hematogenously spread from cutaneous sources. Predisposing factors including pregnancy, trauma, endocarditis, intravenous drug use, and immunosuppression are usually discovered. Because blood circulation is sluggish in the ilium, infections in this area may begin as osteomyelitis and then extend into the sacroiliac joint [3]. However, in the present case CT and MRI of the lumbar spine were negative. The multiplicity of the lesions, the rounded sclerotic osteolysis of the greater trochanter and the scattered calcifications in the soft tissue suggested of a low-grade infectious arthritis, such as tuberculosis, brucella or fungal infection [13].

Sacroiliitis occurring early in the course of the disease is usually the hallmark of ankylosing spondylitis [3, 20]. However, a bilateral and symmetric distribution is observed in ankylosing spondylitis, whereas unilateral

abnormalities and the absence of additional manifestations from the spine are most typical in infection, as in the present case. Generally, inflammatory and seronegative arthritis of the sacroiliac joints is usually bilateral. Peripheral joints manifestations and the other stigmata of the diseases including iriditis, uveitis, rashes and bowel dysfunction are usually observed [3, 24, 33].

Osteitis condensans ilii is usually identified on radiographs in young multiparous women following pregnancy. It has been proposed to be the aftermath of ligamentous disruption during pregnancy or parturition. It is a self-limiting condition, rarely leading to chronic sacroiliac joint pain [3]. Metabolic conditions usually affect the sacroiliac joints bilaterally, and are always associated with peripheral manifestations in other joints [3].

Definitive diagnosis is obtained by fine needle aspiration or open biopsy [18, 28]. Acid-fast bacilli in direct smears and stains, the growth of the bacilli in the Löwestein-Jensen culture or the granulomatous lesion identified in the histologic specimen will confirm the diagnosis of tuberculosis. Positive culture results and an antibiogram are highly desirable, because fungal

infection and brucellosis often yield similar histologic findings [24, 32]. However, false negative results should be anticipated in long standing tuberculosis, due to the paucibacillary nature of the disease [34].

Before the advent of chemotherapy, sacroiliac joint arthrodesis was considered of value in hastening the end result, which, regardless of the treatment, was the spontaneous ankylosis of the joint [37]. Kim et al. [21] described two options of operative intervention; joint curettage at the early stages of the disease, or curettage combined with arthrodesis when joint instability is anticipated. Operative results are excellent [20, 34] in terms of residual disease and pain relief.

Currently, multiagent anti-tuberculous chemotherapy is used as the treatment of choice for tuberculosis [36, 39]. In the present patient, the authors elected to treat the sacroiliac joint lesion conservatively and to operate directly on the tuberculous abscess to prevent spreading to the hip joint [27]. The patient had an excellent outcome, according to the healing criteria of Kim et al. [21]. Analogous response to either conservative or surgical treatment has also been reported [2, 6, 11, 20, 21, 24, 34, 37, 38]. Recurrences are generally not anticipated [21, 34].

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