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Casi clinici

Case reports

Tuberculous sacro-ileitis: two cases and radiological findings

Sacroileite tubercolare: descrizione di due casi e aspetti radiologici

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INTRODUCTION

Musculoskeletal system involvement in tuberculosis (TB) is observed in 1-5% of cases and the sacroiliac (S) joint is involved in 3-9.7% [1-3]. Tuberculous sacroileitis is rare and generally an isolated phenomen, and should receive early diagnosis for a good prognosis [4, 5].

Tuberculous spondylitis involving the lower thoracic and upper lumbar vertebrae (31%) and unilateral sacroileitis(19%) are less commonly observed [6, 7].

Brucellosis is frequently involved in sacroiliac joints [1]. In general, patients with osteoarticular tuberculosis have a long duration of symptoms of the disease prior to the diagnosis (median: 5.5 months) [1, 7].

Computed tomography is very useful in detecting early involvement of the sacroiliac joints and in defining the extent of the abscesses and the severity of the involvement in patients with spondylitis [2, 4, 5, 7].

Tuberculosis of the bones and joints is an elusive diagnosis [8, 9].

The sacroiliac joint is rarely affected; only about 15 articles on TB sacroileitis (TBS) have been published over the last 20 years [1, 10]. We report two cases of TBS with details of radiological studies and review of the literature concerning TBS [9-12]. The current diagnosis and treatment of this condition is discussed based on these cases and a literature review.

CASE REPORTS

Case 1: A 26-year-old female who was treated for pulmonary TB four years ago, visited our outpatient unit with one month history of progressive pain at her right buttock. Recently she had had pain also at rest and frequently awakened at night due to pain. The patient had sporadic systemic episodes of low-grade fever and night sweats. Physical examination revealed a slight right-sided antalgic gait, and there were no reflex, sensory, or motor changes in either lower limb. Faber's test (flexion, abduction, external rotation) was positive for the right hip.

The sacroiliac joint was tender in deep palpation and stretching manoeuvre, including lateral pelvic compression test, Patrick and Gaenslen's tests. In laboratory examination, complete blood count, routine biochemical and serologic tests including rheumatoid factor were all normal, and standard brucella agglutination (STA ≥1/160) test was negative.

Erythrocyte sedimentation rate (ESR) was 42 mm/h, and C-reactive protein (CRP) was 4.12 mg/dl (normal 0-0.8). A tuberculin skin test was positive, with an induration of 18 mm. Anteroposterior radiography of the pelvis revealed sclerosis, and blurring of the joint margin of the right sacroiliac joint (Figure 1). In chest x-ray, there was bilateral pulmonary apex thickening which was consistent with an old tuberculosis image. Magnetic resonance imaging (MRI) methods from osseous window demon-

Figure 1 - Anteroposterior radiography of the pelvis revealed sclerosis, and blurring of the joint margin of the right sacroiliac joint.

Figure 2 - Computed tomography (CT) of the pelvis showed a left sacroiliac joint lesion and significant erosions, in addition to scattered calcifications and extensive destruction of the sacroiliac joint areas.

strated increased liquid in iliac osseous of right sacrum. Godolinium-enhanced axial MRI of sacroiliac joints showed decreased T1 and increased T2 signal in the right iliac and lateral sacrum suggestive of sacroileitis possibly of infectious origin. Also in the right muscle of piriformis, there was a large soft tissue mass (short axis 6 mm) at the right hind-thigh, with signal abnormalities suggesting an abscess. Aspiration biopsy of the abscess showed epithelioid and giant-cell granulomas with caseous necrosis; culturing on Loweinstein-Jensen medium recovered the tubercule bacillus. The patient was administered a four anti-tuberculous chemotherapy regimen including isoniazid (15 mg/kg body weight), rifampicin (10 mg/kg body weight), ethambutole (15 mg/ kg body weight), and pyrazinamide (35 mg/kg body weight) for 2 months followed by double anti-tuberculous (isoniazid plus rifampicin) therapy for ten months. Within the last year, clinical symptoms resolved completely and MRI showed complete regression of the abscess.

Case 2: A 24-year-old healthy male was admitted to the University Hospital with severe lower back pain that radiated to the left buttock, thigh and leg for two months. He denied lower extremity weakness, numbness, paresthesias, or urinary, or bowel incontinence. He had no recent history of weight loss, night sweats, altered bowel habits or urethral discharge. In laboratory examination, complete blood count, routine biochemical and serologic tests including rheumatoid factor were all normal, and standard brucella agglutination test (STA titer 1:80) was negative. Erythrocyte sedimentation rate was 65 mm/h and CRP was 0.9 mg/dl (normal value <0.5). There was a mild hypochromic and microcytic anemia, hematocrit was 39.5%. Tuberculin skin test was positive (14 mm); however the patient had a history of BCG vaccination 10 years previously. He was treated for four weeks with empiric antibiotics for brucellosis (rifampicine 600 mg/day plus doxycycline 200 mg/day). However, the clinical course did not ameliorate. Plain antero-posterior radiograph of the pelvis showed a left sacroiliac joint lesion and significant destruction and sclerosis. Computed tomography (CT) of the pelvis showed a left sacroiliac joint lesion and significant erosions, in addition to scattered calcifications and extensive destruction of the sacroiliac joint re-

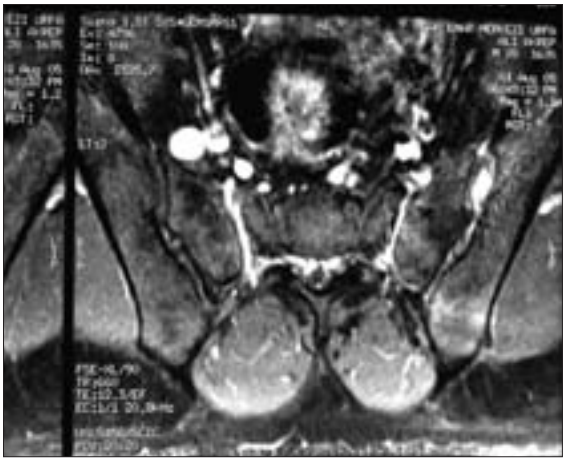

Figure 3 - MRI of sacroiliac joints showed decreased T1 and increased T2 signal in the right iliac and lateral sacrum suggestive of sacroiliitis possibly of infectious origin.

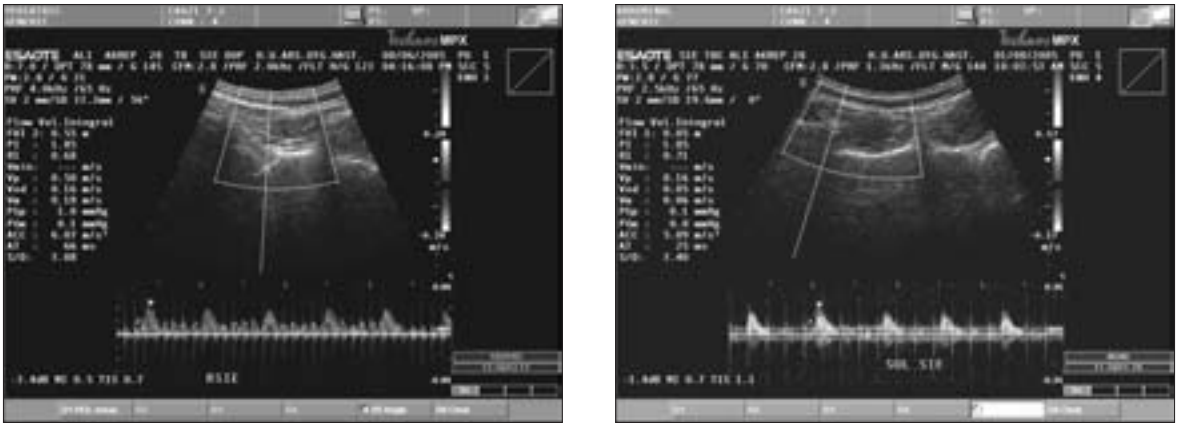

Figure 4 - Colours Doppler ultrasonography (CDUS) of sacroiliac joints. Resistive Index (RI): right 0.68 and left 0.71 which defined inflammation sacroiliac areas.

gion (Figure 2). Chest x-ray, CT and MRI of the lumbar spine were unremarkable. Magnetic resonance imaging methods from osseous window demonstrated left sacrum oedema and increased liquid in iliac osseous. Godolinium-enhanced axial MRI of sacroiliac joints showed decreased T1 and increased T2 signal in the right iliac and lateral sacrum, suggestive of sacroileitis possibly of infectious origin (Figure 3). Technecium-99 three-phase bone scintigraphy demonstrated increased radioisotope uptake at the left sacroiliac joint. Colour Doppler Ultrasonography (CDUS) of sacroiliac joints demonstrated the resistive index (RI) to be 0.68 in the right and 0.71 in the left, which defined inflammation in the sacroiliac regions [12] (Figure 4). We performed CT-scan-guided needle aspiration and biopsy from left sacroiliac joint for diagnosis. Direct smear and staining of the material showed acid-fast bacilli. Histological examination was consistent with a granulomatous infection. TB bacillus was isolated in Lowenstein-Jansen culture medium. The patient was administered a four anti-tuberculous chemotherapy regimen including isoniazid (15 mg/kg body weight), rifampicin (10 mg/kg body weight), ethambutole (15 mg/ kg body weight), and pyrazinamide (35 mg/kg body weight) for 2 months, followed by double anti-tuberculous (isoniazid plus rifampicin) therapy for ten months. They experienced significant improvement soon after the initiation of antituberculous therapy. On follow-up visits four months later, control MRI of the sacroiliac joints showed improvement, ESR and CRP levels had decreased to normal levels and he was almost symptom-free.

DISCUSSION

The clinical symptoms of tuberculous sacroileitis are buttock and lower back pain in most patients, and most have difficulty in walking, and have radicular pain in their lower limbs. Diagnosis is usually delayed, because of the nonspecific clinical features and the difficulty to explore the sacroiliac joint [1, 10,11]. In our region brucella is hyper endemic and sacroiliac joint involvement is frequently observed. However buttock or lower back pain is noted also for brucellar sacroiliac joints involvement. Clinical findings can mimick brucellar or TBS joints involvement. Also medical treatment of TBS is often sufficient [4-7]. The authors classified TBS into four types based on the clinical and radiologic findings [10]. Types 1-2 are treated conservatively with chemotherapy alone, wheras types 3 and 4 are treated with surgery and chemotherapy [1, 2, 4]. Scan abnormalities directed clinicians towards appropriate further workup and diagnosis of unilateral TBS [8, 11]. Sacrocoxalgia is accompanied in 50% of cases by another focus of active tuberculosis [2]. Haemotological data are frequently noncontributory, but higher ESR and positive skin reaction may be very useful. Open biopsy, histology, and cultures establish the diagnosis of tuberculosis [6]. Accurate diagnosis is based on a percutaneous synovial fluid or abscess aspirate bacteriology. CT-scan, MRI, and radionucleid bone scanning and CDUS are the most helpful radiological examinations [7, 12, 13]. Nuclear bone scanning should be kept in mind when assessing spinal pain in patients with a high risk

of tuberculosis infection or reactivation [8, 11]. Its insidious presentation leads to delayed diagnosis [6]. Constitutional symptoms are typically subtle or absent in extrapulmonary TB. The reported prevalence of pulmonary TB concurrent with osteo-articular TB is approximately 50%, but our second patient had no other focus of the disease. The pathogenesis of our patient's disease could not be determined; however osteo-articular TB is thought to result from reactivation of latent foci seeded during the primary illness or to occur secondarily to hematogenous or lymphatic spread of *M. tuberculosis* from reactivated pulmonary or extrapulmonary foci, such as in our first patient [1, 7, 13]. The absence of another focus in the second patient is consistent with its infrequent association with osteoarticular TB [14].

Plain radiograph may not show any abnormality in the early stage of TBS [5, 13]. Subsequently, haziness and/or loss of the joint line, irregularity of the articular surface and subchondral erosions appear [2, 7, 10]. In later stages, descructive lesions become more distinct and cavitation develops [7, 13]. Fusion of the sacro-iliac joint can occur within 3-4 years of onset of medical treatment [10, 13]. Bone scan is helpful for early recognition of the condition; however, CT or MRI are more helpful for early detection of sacroiliac tuberculosis [1, 3, 9, 11]. CT scan shows clearly the extent of joint destruction and MRI delineates the abscess in the soft tissues. MRI may also contribute to the differential diagnosis from a soft tissue tumor or pyogenic arthritis [7, 8, 11, 13].

Tuberculous sacroileitis should be differentiated from degenerative and post-traumatic arthritis; pyogenic infection of the sacroiliac joint; inflammatory disease such as sero-negative spondyloarthropathies or brucellar arthritis, ankylosing spondylitis, psoriatic arthritis, Reiter's and Behçet's syndromes and inflammatory bowel diseases; connective tissue disorders such as rheumatoid arthiritis and systemic lupus erythematous; familial Mediterranean fever with multisystem involvement; arthritis in pseudogout and hyperparathyroidism; tumors and tumor-like conditions, and pigmented villonodular synovitis [1, 2, 5, 9, 11]. The tuberculin skin test is usually positive in skeletal TB. The diagnosis is made by isolation of mycobacteria either by acid fast bacilli stain or by culture [9]. We employed CT-scan-guided needle aspiration [1]. Slowly developing, indolent involvement of the bone, joint or soft tissues may delay diagnosis for months to years [10, 11]. Currently multiagent anti-tuberculous chemotherapy is the treatment of choice for tuberculosis. The patients have an excellent outcome, according to the healing criteria of Kim et al. [10]. Sacro-iliac joint is diffucult to explore and has recently beneficiated from technical improvement in imaging and medical treatment of tuberculosis sacroilitis is often sufficient.

In conclusion, early recognition of TBS is extremely difficult, due mostly to the insidious nature of the disease and the lack of clinical evidence of the disease at the time of initial evaluation. The sacroiliac joints and back pain must be considered, particularly in patients with severe buttock pain or in case of a suspicious appearance of sacroileitis on plain imaging methods (CT and MRI), and Technecium-99 three-phase bone scintigraphy. This presentation suggests that if sacroiliac joints and back pain persists in seronegative brucellosis despite appropriate treatment, the possibility of sacroiliac joints disease such as TBS should always be considered especially in endemic regions for brucellosis.

Key words: Tuberculous sacroileitis, abscess, magnetic resonance imaging, treatment, low back pain

SUMMARY

Infective sacro-ileitis is due to common bacteria, 25% being tuberculosis and 10% brucellosis. Slow progression characterizes joint tuberculosis, an uncommon variant of this disease. The onset is usually insidious, and early diagnosis requires a high index of clinical suspicion.

We report two cases with tuberculous sacro-ileitis which initially mimicked brucellosis infiltration. Diagnosis of tuberculosis of the sacroiliac joint was established by fine-needle aspiration of joint and radiological imaging methods such as computerized tomography, magnetic resonance and three-phase bone scan.

The current diagnosis and treatment of this condition is discussed based on these cases and a literature review.

RIASSUNTO

La sacro-ileite infettiva riconosce la propria eziologia in batteri di usuale riscontro, ma micobatteri e brucelle possono essere isolati nel 25% e nel 10% dei casi, rispettivamente. La tubercolosi articolare, una variante poco frequente di questa patologia, è caratterizzata da una lenta progressione.
L'esordio è in genere subdolo, e la diagnosi precoce richiede un elevato indice di sospetto clinico. Nel presente articolo riportiamo due casi di sacro-ileite tubercolare che ha inizialmente simulato un'infiltrazione brucellare. La diagnosi di tubercolosi localizzata all'articolazione sacro-iliaca è stata posta avvalendosi dell'aspirazione con ago sottile e delle metodiche per immagine, quali la tomografia computerizzata, la risonanza magnetica e lo scan osseo trifasico. Verranno discussi le attuali procedure diagnostiche e il trattamento di tale patologia basandosi sui casi descritti e sulla revisione della letteratura.

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