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Case Report Open Access

Disabling Bilateral Hip Disease and Femoral Bone Defects Revealing Systemic Sarcoidosis

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Abstract

Introduction: Bone involvement in sarcoidosis is rare and often asymptomatic. The small bones of hands and feet are the most common localizations, while skull, knee, rib, pelvic and sternal localizations are rarely reported. Here we report a hip localization.

Case: A 52-year-old woman consulted for acute bilateral coxopathy. Chest radiography objectified bilateral interstitial syndrome with mediastinal lymphadenopathy. The X-ray of the pelvis and both hips showed no abnormalities and CT scan revealed a moth-eaten osteolytic lesion of the left femoral head. It was associated with inflammatory syndrome and cholestasis. Liver biopsy revealed epithelioid and giant cell granulomas without caseous necrosis. The pathology specimens were interpreted as representing sarcoid.

Conclusion: Well, this observation of systemic sarcoidosis, authenticated by histological evidence, seems original mostly because of its revealing clinical presentation of the disease, with violent bilateral coxopathy *and* total functional disability, but also radiological, with an aspect of moth-eaten osteolysis of the femoral head. No similar cases have been reported in the literature.

Keywords: Sarcoidosis; Hip bone; Magnetic resonance imaging

Introduction

Sarcoidosis is a systemic granulomatous disorder. Although it usually presents with bilateral hilar lymphadenopathy and lung infiltration, multiple organ systems may also be affected. Bone involvement is quite rare (5% to 10%) and mostly limited in small tubular bones of the hands and feet. In addition, it is usually occurred in patients with chronic course or known multisystem sarcoidosis and it is often asymptomatic [1,2]. Here we report an original case of systemic sarcoidosis, pulmonary, liver and bone revealed by violent bilateral buttock and groin pain and moth-eaten osteolysis of the left femoral head.

Case Report

A 52-year-old woman presented with acute onset of bilateral buttock and groin pain with total functional disability, she was unable to walk because the pain was severe. Physical examination found pain with limitation of both hips and it was thought that a coxopathy was the probable cause. X-ray of the entire pelvis and both hips showed no abnormalities. Laboratory tests showed inflammatory syndrome (C-reactive protein=41 mg/L, erythrocyte sedimentation rate=67 mm the 1st hour), cholestasis (3 times normal) without cytolysis. Complete blood counts showed leukopenia (white blood cells=3300 cells/mm), blood electrolytes were normal. Serum calcium was 2 mmol/L and 4 mmol/L. Radiographs of the pelvis failed to show any osseous abnormality to suggest an aggressive lesion (Figure l).

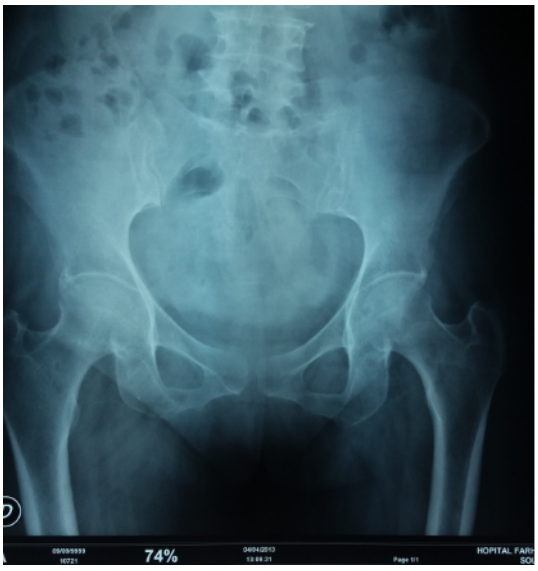

Figure 1: X-ray of the entire pelvis and both hips showed no abnormalities.

Chest radiography objectified bilateral interstitial syndrome with mediastinal widening; thoraco-abdominal pelvic CT scan confirmed the presence of diffuse reticulonodular images in both lung fields with

large mediastinal nodes and showed splenomegaly. CT scans of the pelvis showed moth-eaten osteolysis of the left femoral head (Figure 2).

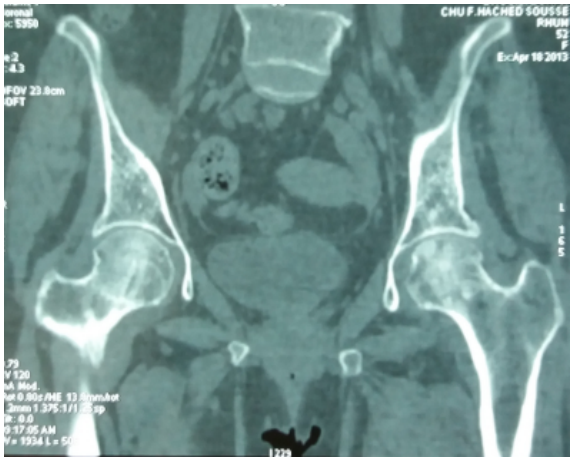

Figure 2: CT scans of the pelvis showed moth-eaten osteolysis of the left femoral head.

Further workup with 99mTc whole-body bone scintigraphy revealed mildly increased activity within both femoral heads and posterior arch of the 5th and 7th right ribs. No other skeletal lesions were identified. MR imaging of the pelvis was ordered for further evaluation. It showed multiple low-signal-intensity lesions were seen throughout the head and the femoral neck on TI-weighted MR images (Figure 3); the lesions became mildly hyperintense on T2-weighted images (Figure 4). These lesions showed inhomogeneous enhancement after intra venous gadolinium administration (Figure 5).

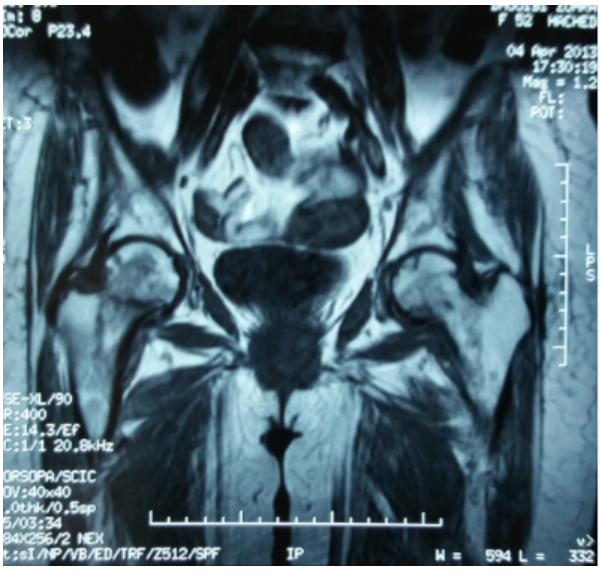

Figure 3: T1-weighted MR image shows multiple low-signal-intensity lesions were seen throughout the head and the femoral neck.

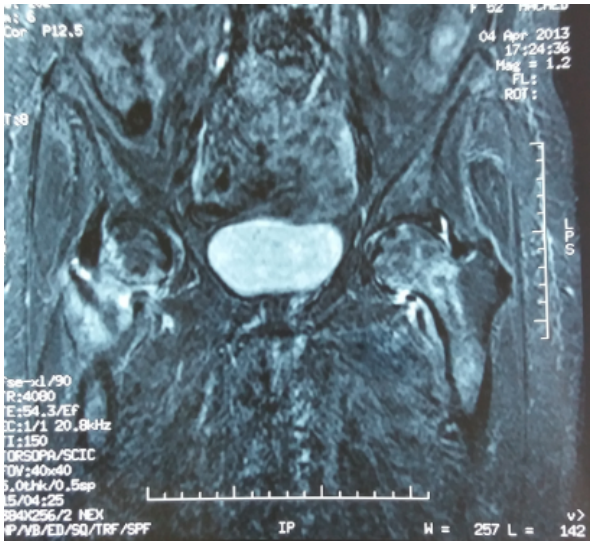

Figure 4: T2-weighted MR image shows the lesions to be mildly hyperintense.

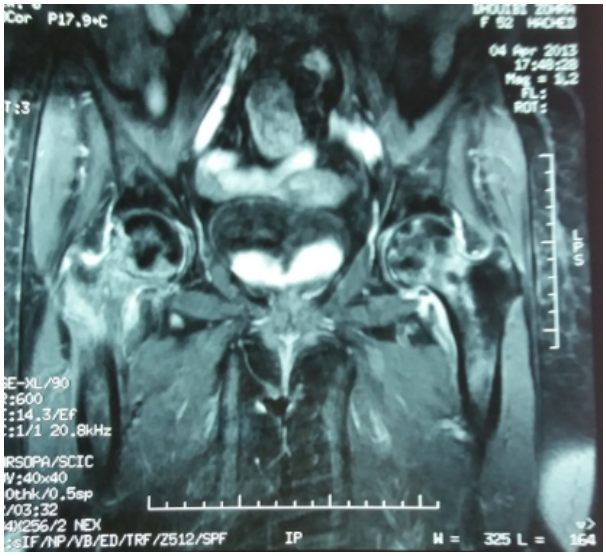

Figure 5: Gadolinium-enhanced T1-weighted MR image reveals inhomogeneous enhancement after intra venous gadolinium administration.

The MR imaging findings raised concern for lymphoma, or even sarcoidosis, given the patient's history. A salivary glands biopsy and myelogram failed to show any granulomatous or neoplastic abnormality. Consequently, a liver biopsy was performed. The pathology report indicated epithelioid and giant cell granulomas without caseous necrosis. The bronchoalveolar lavage revealed lymphocytic alveolitis (40 × 10 cells with 40% lymphocytes, the reste were macrophages) without Mycobacterium tuberculosis on direct examination and culture.

The patient's pathology slides and MR images were sent to a tertiary referral center for consultation. The pathology specimens were interpreted as representing sarcoid. The patient had a complete and spontaneous regression of bone and joint symptoms after one month of evolution. Having liver damage, we began corticosteroid therapy (0.5 mg/kg) with a favorable outcome. The patient remained asymptomatic at a 3-year follow-up.

Discussion

Bone lesions are fairly uncommon in sarcoidosis (5% to 10% of cases). It predominantly affects the small tubular bones of the hands and feet. Exceptional locations at the spine [1], long tubular bones, skull, ribs, and pelvis are also reported [2-4]. We report the case of a 52-year-old women in whom systemic sarcoidosis was revealed by bone involvement of the hips. No similar cases have been reported in the literature.

Bone involvement of sarcoidosis is rarely indicative of diagnosis. Indeed, it usually occurred in patients with chronic course or known multisystem sarcoidosis and it was often asymptomatic or caused nonspecific symptomatology such as soft-tissue swelling, pain, and stiffness [2]. In addition, 80% to 90% of patients with skeletal sarcoidosis have concomitant pulmonary abnormalities, whether mediastinal and right paratracheal adenopathy or pulmonary parenchymal disease [5]. In our case, bone involvement of sarcoidosis was revealed by a disabling bilateral hip disease; in addition, concomitant lung disease, both mediastinal adenopathy and pulmonary parenchymal disease, was diagnosed.

The classic osseous lesions within the metacarpals or phalanges of the hands have coarse trabeculations resulting in a lacelike appearance. Other osseous manifestations are well-defined lytic lesions with sclerotic margins, aggressive-appearing lytic lesions, and, only infrequently, osteosclerosis [3,6]. In our case, it was a moth-eaten osteolysis.

Well, this observation of systemic sarcoidosis, authenticated by histological evidence, seems original mostly because of its revealing clinical presentation of the disease, with violent bilateral coxopathy and total functional disability, but also radiological, with an aspect of moth-eaten osteolysis of the femoral head.

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