SACROILIITIS AS PRESENTATION OF SYSTEMIC DISEASE (Two Case reports)

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Introduction

Sacroiliitis may sometimes be the presenting feature of an acute systemic illness. The diagnosis may be overlooked due to lack of awareness of this infrequent manifestation and to poorly localised presenting symptoms [1]. As aspiration of the sacroiliac joint is often very painful and needs expertise and a plain radiograph rarely proves helpful in the early stages, it is the report of an early blood culture, serology where indicated, combined with radionuclide scanning that clinches an early diagnosis and its localisation. It is important to be aware of this form of presentation as timely intervention will result in complete resolution without a crippling arthropathy [1,2]. We report two systemic infectious diseases presenting primarily with features of sacroiliitis.

Case 1:

A 35-year-old serving soldier presented with six weeks history of progressive general ill-health, occasional low grade fever, easy fatigability and pain in the right hip which worsened on squatting. The pain was dull aching in nature and radiated down the right leg. He had analgesics on his own without relief and now walked with a limp. On routine physical examination he was found to be mildly febrile (temp 100° F) and had cervical and axillary lymphadenopathy (2-3 cm, multiple, soft, discrete and non tender). Liver and spleen were just palpable. He also had an effusion in the left knee joint that was non tender and had no signs of an active inflammation.

Laboratory investigations revealed a raised ESR (38 mm fall in the first hour by Wintrobe's) and a normocytic normochromic anaemia (Hb 10.0 gm%). The total and differential blood counts were normal. Serum was positive for C-reactive protein and negative for rheumatoid factor. Blood cultures drawn on two occasions

were sterile. A fine needle aspiration of an enlarged cervical lymph node revealed features of reactive hyperplasia. Plain radiographs of the chest and of the left knee were normal. A mantoux test done was negative. Aspiration of the left knee joint yielded straw coloured fluid rich in proteins with predominantly lymphocytes on cytology. Culture of the fluid was sterile. Radiograph of the sacroiliac joints showed early symmetrically destructive bone lesions. A Technitium-99m-methylene diphosphonate imaging was undertaken which showed an increased uptake in both sacroiliac joints (more on the right).

Based on the clinical findings of fever, lymphadenopathy, hepatosplenomegaly and arthritis with history of contact with cattle in his farm at home, a brucella agglutination titer was called for. The standard agglutination test (SAT) for brucella was positive to a titer of 1920; with 2-mercaptoethanol (2ME), the titer was 480 indicating a predominantly IgM response. Based on the result of serology, physical findings and radionuclide imaging a diagnosis of sacroiliitis due to brucellosis was made and specific therapy started with a combination of doxycycline and streptomycin. The patient responded well; bone lesions healed and lymphadenopathy and hepatosplenomegaly regressed over the next three weeks. The agglutination titers remained high over next eight weeks and thereafter showed a slow downward trend. Six months later the Standard agglutination titer was 240 and with 2-ME was 120. The individual was well except for mild residual pain in the right hip. Plain radiographs of the hip joints at follow up after six months showed resolved bone lesions. He remains free of symptoms and without any residual deformity of the affected hip joint.

Case 2:

An 18-year-old male medical student noticed a throbbing pain in the low back region on getting up one morning. By evening he had developed high fever. On examination the temperature was 101° F, he had severe pain over the left buttock and was unable to bear weight on his left leg. Physical examination revealed involvement of the left sacroiliac joint. There was no hepatosplenomegaly or lymphadenopathy. Laboratory investigations revealed a mild polymorphonuclear leucocytosis. Blood cultures taken grew *Sal-*

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monella typhi sensitive to ciprofloxacin, ampicillin, gentamicin, chloramphenicol and cefotaxime. Widal titers showed an increase of agglutination titer of TO (antibody to O antigen of *Salmonella typhi*) to 7,680. An initial plain X ray of the sacroiliac joints did not show any abnormality. However, a radionuclide imaging with Technitium-99-methylene diphosphonate (99mTcMDP) showed hyperactivity in both the blood pool and bone phases in the left sacroiliac joint. The patient was put on intravenous ciprofloxacin along with ampicillin and over next 48 hours exhibited dramatic relief which continued thereafter. The Widal titers rose further during the subsequent week and thereafter started scaling down over next four weeks. Residual pain in left sacroiliac joint lasted for about eight weeks after the patient became afebrile. On regular periodic follow up over next six months the individual remained asymptomatic; the pain in the left hip had gradually disappeared and there was no residual deformity on examination of the affected joint.

Discussion

Brucellosis may often present with arthritis and vague symptoms such as malaise and easy fatigability. In a series of 400 patients in Kuwait, 26% had some form of arthritis [2]. The joints most commonly involved are the sacroiliac joint and knee followed by the hip and spine. Skeletal involvement may be bilateral in about 16.4% cases [3]. Joint effusions have been documented in 1/3 of cases predominantly in the acute group [3]. Our patient also had an effusion in the left knee but his presentation was subacute. Plain radiographs do not show any major pathological changes in the early cases. Bone scintigraphy is more helpful than conventional radiography in early detection of cases. Among the laboratory tests, brucella agglutination titers is a helpful investigation. Blood culture remains an important investigation and is to be collected preferably in Castenada biphasic medium maintaining meticulous asepsis during the procedure. Treatment with a combination of streptomycin plus tetracyclines or rifampicin results in rapid cure rate and resolution of arthritis without sequelae [4]. In subacute cases, pain in the hip joint may sometimes only be elicited by the Gaenslen and FABERE (Flexion, Abduction, External rotation and Extension) manoeuvre [2]. In the case presented, the diagnosis would have been overlooked had the treating physician not been aware of this entity. As brucellosis is endemic in

our country, such awareness is imperative.

Salmonella arthritis presenting as acute pyogenic sacroiliitis is reported in only 0.24% of patients with salmonella infections [5]. In an extensive review of septic sacroiliitis, there were only 4 cases due to salmonella organisms [6]. *Salmonella typhi* has been isolated only rarely [7,8]. Salmonella arthritis presents more commonly as reactive arthritis with migratory symmetrical polyarthropathy 1-3 weeks after a gastrointestinal manifestation [9,10]. Our case is not one of reactive arthritis in view of clinical profile, positive blood culture and swift response to antibiotics. Such a presentation needs to be kept in mind in today's era of changing profile of enteric fever. As blood cultures are positive only in half the cases [1], and joint aspiration may not always be possible, Widal titers may help in early case detection.

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