

## Case Reports

# PONCET'S DISEASE: A REPORT OF FOUR CASES AND REVIEW OF THE LITERATURE

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### ABSTRACT

Four cases of polyarthritis concomitant with active tuberculosis is reported. In three patients pulmonary tuberculosis was confirmed by identification of *Mycobacterium tuberculosis* in the sputum and bronchoalveolar lavage specimens, and in another one tuberculous lymphadenitis was confirmed by excisional biopsy. In all patients arthritis resolved by tuberculosis treatment and did not recur during a follow up period of 18 months to 12 years.

The findings of the presented cases are compatible with Poncet's disease (tuberculous reactive arthritis).

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**Keywords:** Reactive arthritis, Tuberculosis, Poncet's disease.

### INTRODUCTION

Poncet's disease (PD) is described as a polyarticular arthritis which occurs during active visceral tuberculosis (TB) infection.<sup>1</sup> Despite the many case reports<sup>2-13</sup> the concept of PD is not accepted as a specific disease.<sup>7-8</sup> The following four cases of reactive arthritis (ReA) associated with *Mycobacterium tuberculosis* (Mtb) infection present additional information in supporting the existence of PD. These patients attended or referred to Shaheed Beheshti Hospital in Babol.

#### Case 1

An 18-year-old girl presented in September 1980 with arthritis in her ankles, right knee, left elbow and right wrist of three month's duration. In physical examination the knee joint was swollen and tender, the ankle joints were also tender and slightly swollen, and the wrist and elbow joints were painful in palpation and movement. The remainder of the examination was normal. A complete blood count (CBC)

and routine laboratory tests including the latex agglutination test for rheumatoid factor (RF) and antinuclear antibody by immunofluorescence (ANA) were negative. The erythrocyte sedimentation rate (ESR) was 39 mm/hour, and C-reactive protein (CRP) was positive. The radiographs of the wrist and knee joints were unremarkable. Ibuprofen was administered for symptomatic treatment but she did not respond to full doses of ibuprofen during the subsequent visits. Two months later physical examination revealed enlarged cervical lymph nodes which were excised and showed tuberculous lymphadenitis. One month after anti-TB drug therapy the arthritis resolved and did not relapse up to 12 years after discontinuation of the drugs.

#### Case 2

A 14-year-old girl presented with acute polyarthritis involving the right hip and knee joints, left ankles and bilateral elbow joints, and the left wrist for three days in 1988. Over two weeks of observation, the arthritis showed a migratory pattern similar to rheumatic fever. Physical examination showed tenderness, swelling and effusion in the knee joints, movement of the hip joint was very painful and severely limited, and the other joints were also tender and

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swollen. CBC showed a mild leukocytosis, the ESR was 59 mm/h, the CRP was positive, the antistreptolysin O (ASO) titer was 333 Todd units, ANA and RF were negative, and other laboratory tests were unremarkable. Administration of 100 mg/kg aspirin for two weeks was not effective and other non-steroidal anti-inflammatory drugs (NSAIDs) also had limited benefits. A diagnosis of juvenile chronic arthritis was established and during the next six years, arthritis was partially controlled by chloroquine phosphate or sulphasalazine and NSAIDs. During the follow up period, episodes of partial remission and exacerbation were seen. In July 1994, joint symptoms progressed and severe polyarthritis involving bilateral proximal interphalangeal (PIP), metacarpophalangeal (MCP) and metatarsophalangeal (MTP) joints, elbow and knee joints developed. The right hip and both sacroiliac joints were also affected. In physical examination hand joints were severely distended with effusion in most PIP joints. The knees and elbows were tender and swollen, movement of the right hip was painful and limited and sacroiliac joints were tender, aspiration of the knee joints revealed inflammatory fluid which was negative in culture. The ESR was 76 mm/h, CRP was +++, ANA and RF were negative, radiographs of the knees and hands showed soft tissue swelling and radiographs of the pelvis showed normal sacroiliac joint appearance but degenerative changes in the right hip with severe joint space narrowing and subchondral bone sclerosis in the femoral head.

Oral methotrexate 7.5 mg weekly was added. During the next six weeks, joint signs subsided but cough and dyspnea appeared. Chest auscultation revealed wheezes and rhonchi, and the chest radiograph revealed pulmonary parenchymal infiltration. Ziehl Neelson staining of the sputum revealed Mtb.

Antirheumatic drugs were discontinued and anti-TB therapy was substituted. Joint symptoms and signs subsided completely after six weeks and did not recur up to five years after treatment of TB.

#### Case 3

In March 1995, A 33-year-old woman who had been well previously presented with arthritis of the left ankle, left shoulder and right wrist, pain in the right forearm, and the appearance of erythema nodosum on the anterior surfaces of the tibia bilaterally over a two weeks period. In physical examination the ankle joint was severely tender and slightly swollen. There were several tender, red and erythematous nodules on the tibial surfaces and the results of clinical examination in the remainder of the musculoskeletal system were unremarkable. Routine blood tests including CBC, RF, ASO, ANA and CRP were negative. The ESR was 19 mm/h, and the chest radiograph showed left hilar adenopathy. Pulmonary and mediastinal CT scanning revealed multiple bilateral adenopathies in both hilar and para-aortic areas. The specimen obtained by fiberoptic bronchoscopic alveo-

lar lavage revealed Mtb on smears and culture. Arthritis subsided after six weeks of anti-TB treatment and did not relapse up to three years later.

#### Case 4

A 49-year-old man was admitted in January 1997 with fever, weakness, arthralgia and arthritis. The temperature ranged between 37.5 -39°C over two weeks of hospitalization. There was tenderness and slight swelling in both ankles and mild localized edema on the dorsal regions of the tarsal joints bilaterally and moderate difficulty in walking. The knee joints were slightly tender. Blood count, urine culture, urinalysis, blood cultures and liver function test were normal. RF, ANA, ASO, Coombs wright agglutination and PPD test were negative. Other febrile illnesses were ruled out by appropriate tests. Chest radiograph and CT scanning revealed bilateral hilar adenopathies. Fiberoptic bronchoscopic study showed no endobronchial lesion. The specimen obtained by bronchoalveolar lavage and the sputum staining and cultures revealed Mtb. Fever and joint pain subsided two weeks later without any treatment, but swelling and edema disappeared after two months of anti-TB therapy. The joint signs did not recur over an 18 month follow up period.

### DISCUSSION

Four cases of simultaneous ReA and active TB infection are reported here. In three patients pulmonary TB was confirmed by bacteriologic studies and in the fourth case, TB lymphadenitis was established by histologic examination of the lymph node. The diagnosis of the presented cases, regarding the pattern of joint involvement affecting predominantly the large joints of the lower limbs and the rapid response of arthritis to antituberculous therapy is consistent with tuberculous ReA (PD). The diagnosis of PD remains clinical and is established on excluding other potential causes of arthritis in a patient with active TB.<sup>3</sup> The complete resolution of arthritis symptoms on anti-TB therapy also furnishes further proof of the diagnosis.

Involvement of the musculoskeletal system, such as spondylitis, osteomyelitis or arthritis occur in only 2% of patients with TB<sup>14</sup> but should be considered in any patient with rheumatic symptoms. Unlike some reported cases<sup>2,4,5</sup> with direct involvement of bone by Mtb, the presented cases show no evidence of direct skeletal involvement. The time from the onset of arthritis to diagnosis of TB in this study ranged from two weeks to six years. In other reported cases disease duration ranged from two weeks up to one year,<sup>3,5,7,11,12,15</sup> but cases with a prolonged disease course similar to our second patient have not been reported up to now. The clinical picture of this patient in particular when she presented with symmetric polyarthritis was indistinguishable from rheumatoid arthritis (RA); however, with regard to the rapid response of arthritis to anti-TB treatment, RA

was excluded and PD was confirmed.

A rheumatoid arthritis-like presentation with symmetric involvement of small finger joints accompanying bacteriologically confirmed TB has been reported by Dlugovitzky et al.<sup>16</sup> Three out of the five patients remained arthritic by the time of bacteriologic conversion and fulfilled the criteria for RA. In the two remaining patients sputum negativization was accompanied by disappearance of rheumatic manifestations in favour of PD.

PD is an entity characterized by ReA developing in the presence of active TB elsewhere. Any joint can be affected, however the knees, ankles and elbows are involved more often than small joints of the hands and feet.<sup>17</sup>

ReA is mediated by an immune response against whole bacteria or their fragments, which is carried to the joint.<sup>18,19</sup> The responsible antigen may be HLA-B27 complexed with peptides derived from proteins of arthritis causing bacteria such as heat shock protein (hsp) family.<sup>19,20</sup> The hsp, a ubiquitous protein in a wide range of species from bacteria to mammals, is strongly immunogenic<sup>20</sup> and can induce autoimmune disease.<sup>20,21</sup> *Mycobacterium tuberculosis* (Mtb) antigen in adjuvant arthritis is an hsp<sup>22</sup> which can cross react with cartilage proteoglycan and induce autoimmunity and result in cytokine secretion by CD4 lymphocytes which has been implicated in the pathogenesis of autoimmune arthritis.<sup>11,23-25</sup> Intra-articular administration of recombinant Mtb hsp can induce joint inflammation in Mtb sensitized recipients.<sup>26</sup>

In conclusion, the immune response against Mtb hsp in the genetically predisposed host may explain the mechanism of ReA in PD. The pathogenic similarity of PD and erythema nodosum<sup>3</sup> which is applicable clinically to the third case of the present study has been demonstrated in a case report.<sup>15</sup> Further investigation is required to elucidate the relationship between active TB and ReA.

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