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Case Presentation

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Poncet's Disease - A Commonly Missed Diagnosis

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1. Abstract

One of the rare presentations of both active pulmonary and extrapulmonary tuberculosis is polyarthropathy in the form of involvement of multiple large and small joints in the body. This reactive arthritis in tuberculosis (TB) is known as Poncet's disease, a rare aseptic form of arthritis characterized by polyarticular impairment observed in patients with active TB, without any evidence of direct bacillary invasion of the joints. It is a different entity from tuberculous arthritis, which is usually monoarticular and is caused by direct tuberculin infection. Poncet's disease remains a diagnosis of exclusion. There are very limited case reports even from countries where tuberculosis is common and there are no accepted diagnostic criteria for Poncet's disease. This diagnostic possibility becomes increasingly important as the use of corticosteroids, immune suppressants or biologicals can risk further dissemination of the disease. We describe the case of a 50-year-old woman, who presented with active tuberculosis where polyarthralgia was the first and only symptom for four months. Polyarthritis workup was unremarkable and symptoms were not relieved by non-steroidal anti-inflammatory drugs, but had complete resolution of symptoms after 6 weeks of anti-tubercular therapy. The total duration of therapy was 6 months.

2. Introduction

Tuberculosis (TB) is a major public health concern. It remains the leading cause of mortality attributed to infectious diseases with estimated 1.5 million deaths from TB in 2018-19. Poncet's disease (PD) is a rare syndrome first described in 1897 by the Frenchman Antonin Poncet, when he described a polyarthritis in an acute stage of TB, which resolved without joint damage. Continuous reports on patients with similar characteristics led authors to improve the definition, and in 1978, Bloxham and Addy defined PD as a para infective arthritis. Even when its existence has been questioned by many, cases have been continued to be reported over the years. Poncet's disease is characterized by articular affection in patients diagnosed with TB, not related to direct invasion by the micro-organism, but to an immune reaction to the tuberculous protein, constituting a reactive arthritis [1, 2]. The condition is different from tuberculous arthritis which is usually monoarticular and is caused by direct tubercular involvement of the joint. This may sometimes be the sole manifestation of the disease before more obvious features develop. The pain experienced during polyarthritis can be crippling thereby limiting the mobility and activities of patients. Polyarthritis as a symptom of active tuberculosis can be easily misinterpreted for more common causes of polyarthritis such as rheumatological diseases that have similar presentation.

3. Case Presentation

A 50-years-old female patient reported to the department with painful knees, wrists and shoulder joints since the past 6 months. She also reported gradual deterioration over preceding months, resulting in slight difficulty in walking. The joint symptoms were present throughout the day with no diurnal variation. The intensity of pain in the larger joints including her upper lower limbs, elbow and knee joints has increased over period of time and got aggravated by joint movement and relieved by taking rest and NSAIDs. Patient also gave a history of morning stiffness which

usually lasted for half an hour. She also reported about the low-grade evening rise of temperature, loss of appetite and weight and productive cough with sputum during the same period. There was no history of photosensitivity, oral ulceration, back pain, rash over the body, diarrhea, or burning micturition There was no history of trauma, conjunctivitis, bowel or bladder symptoms, or similar episodes in the past. On examination, pallor was present and there was involvement of both wrist joints, interphalangeal joints and ankle joints in form of pain, tenderness and mild local swelling and there was no lymphadenopathy or erythema nodosum. Investigations revealed TLC-7700 cells/cmm (N-71, L-24, E-01, M-04), Hemoglobin of 9 gm% and microcytic hypochromic picture on peripheral smear. Aerobic throat swab culture was negative. An ESR of 120 mmHg per 1st hour and Mantoux test was strongly positive (20 x 12 mm). Rheumatoid factor, anti-CCP (citrullinated peptide antibodies antibodies), c-ANCA and p-ANCA, anti-nuclear antibody (ANA) and Anti-Streptolysin O (ASLO) titre were negative. USG of Whole Abdomen was normal except for cholelithiasis. Other investigations included Serum CRP (3 mg/L), LDH (204.71 U/L), Uric Acid (4.6 mg%) and ACE (33.9 U/L), LFT(Total Bilirubin-0.3 mg%, Direct bilirubin-0.2 mg%,SGOT-20 U/L,SGPT – 9 U/L, Alkaline Phosphatase – 82 U/L), FBS-149 mg%. PPBS- 86 mg%, Serum Sodium-138 mmol/L, serum potassium-4.3 mmol/L, serum urea-14 mg/dl, creatinine-0.5 mg/dl, all levels were within normal limits. Urine routine examination was normal. Chest X-ray revealed haziness in left mid and lower zones. X-rays of the involved joints showed periarticular soft tissue swelling, and there were no changes of active tuberculosis. Patient's symptoms were not relieved by non-steroidal anti-inflammatory drugs, but had complete resolution of symptoms after 6 weeks of anti-tubercular therapy. The total duration of therapy was 6 months.

4. Discussion

Tuberculosis (TB) is a major public health problem in India. The country has the largest number of TB cases in the world accounting for over a quarter of the global TB and multidrug-resistant TB (MDR-TB) burden. In 2016, 2.79 million people became ill from TB, and 435,000 died from it. India has the greatest number of new cases of MDR-TB (including rifampicin resistance), with an estimated 147,000 cases in 2016. There are more than 850,000 cases of TB each year in India that are either undetected and untreated or diagnosed and treated by healthcare providers with potentially substandard drugs and treatment regimens. Approximately 10% to 11% of the extrapulmonary tuberculosis cases affect bones and joints, corresponding to 1% to 3% of all cases of tuberculosis. The potential of tuberculosis, even when subclinical, to trigger reactive conditions should be remembered. That possibility becomes increasingly important as the use of corticosteroids, immune suppressants or biologicals can trigger the reactivation or dissemination of the disease. It is widely known that tubercular septic monoarthritis, in which M. tuberculosis may be isolated from the joint, may complicate tuberculous infection; however active TB may be complicated by a sterile reactive arthritis is less known and therefore often missed. Poncet's disease is used to indicate an aseptic polyarthritis, presumably a reactive arthritis, developing in the presence of active TB elsewhere.3Although Poncet's disease is considered a reactive arthritis, the clinical presentation of Poncet's disease differs from the classical pattern of reactive arthritis [3, 4, 5].

In contrast to the usual tuberculous arthritis which is monoarticular, infectious and destructive, tuberculous rheumatism or Poncet's disease is a non-destructive para- infective symmetric polyarthritis occurring in patients with active visceral or disseminated tuberculosis, in which there is neither evidence of bacteriological involvement of joint themselves nor any other known cause of polyarthritis detected [5, 6].

Although Poncet's disease is considered a Reactive arthritis, the clinical presentation of Poncet's disease differs from the classical pattern of Reactive arthritis. In contrast to Reactive arthritis, the onset of symptoms in Poncet's disease occur before the start of arthritis and is much longer than just a few weeks, whereas resolution of arthritis upon starting of adequate anti-tuberculous therapy is mostly within a few weeks [7]. Chronic arthritis has never been reported in Poncet's disease. Furthermore, Poncet's disease is generally, not associated with sacroiliitis.

There are various hypothesis put forward to explain the pathogenesis of tubercular rheumatism. Genetic theory has been put forward describing an HLA linked hyper responsiveness to Mycobacterium antigen [8]. Immunological explanation has been given on the basis of finding a hypersensitive immune response to tuberculoprotein [9]. Cross reactive immune response has also been thought of, on the basis of finding antigenic similarity between human cartilage and fraction of Mycobacterium tuberculosis [10].

The tubercular bacilli have been found to be arthritogenic. This fact is based on observing chronic synovitis in animals injected with heat killed dessicated TB bacilli [11].

It has been hypothesized that after infection, as a result of systemic immunization, sensitized CD4+ cells together with bacterial antigens migrate to the joints and cause arthritis. This hypothesis is supported by the animal model of adjuvant arthritis in which injection of heat-killed desiccated M. Tuberculosis (complete Freund's adjuvant) results in arthritis. The human counterpart of this model is observed in patients with bladder cancer receiving immunotherapy by means of intravesical instillation of attenuated M. bacillus Calmette-Guerin. In 0.5% of these patients, a reactive polyarthritis is observed [12].

The mechanism of pain has been attributed to rapid accumulation of periarticular fluid possibly due to exaggerated hypersensitivity

reaction [13].

The joint symptoms may precede other manifestations of tuberculosis or there could be simultaneously involvement of joints with pulmonary tuberculosis.

In a review series of 50 cases, twenty-nine cases (68%) involved male patients. Although by definition, Poncet's disease is a non-septic arthritis, only in 15 cases (30%) was septic tuberculous arthritis ruled out by culture or histology. In five patients (10%)], septic tuberculous arthritis of one joint was demonstrated in addition to presumed non-septic arthritis of other joints. Septic tuberculous arthritis was demonstrated either by histology, culture or characteristic X-ray findings. In two of these patients, septic polyarticular tuberculous arthritis was ruled out by culture, suggesting that these cases represent a mixture of septic tuberculous arthritis and Poncet's disease. In the other three patients no additional joint cultures were performed. Besides the three patients with characteristic X-ray findings due to tuberculous destruction of bone and joints, no irreversible joint damage was observed in the 50 case reports. Fifteen patients (30%) presented with an oligoarthritis of less than four joints and the other patients presented with polyarthritis. In eight patients (16%), no localization of polyarthritis was described. In the other 42 cases, knees (62%) and ankles (57%) were the most commonly involved joints followed by the wrists (48%). In 12 patients (29%), small joints of hand or feet were involved as well. In 10 (20%) patients, the diagnosis of TB was based on the clinical picture only. In 48% of the patients, the TB was localized extrapulmonary. In 50% of the patients, abnormalities on standard chest X-rays were observed.14.6% of the patients presented with erythema nodosum. Resolution of the arthritis with anti-TB drugs ranged from 1 week to 4 months [14].

5. Conclusion

Based on our observations, we propose that tuberculosis be included among the differentials for patients with unusual presentation of joint pains, especially in endemic regions and/or susceptible populations. It is a diagnosis of exclusion but must be included in differential diagnosis especially in countries/regions where prevalence of M. tuberculosis infection is high.

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