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Wind-swept pelvis in poncet's disease: A rare case report

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Abstract

Introduction: Windswept pelvis is a rare feature seen in Poncet's Disease. This rare entity is found in cases with long standing disease. One hip is flexed, adducted and internal rotated while the contra lateral hip is flexed, abducted and external Rotated. Windswept hip deformity may occur in association with hip dislocation and scoliosis. Children with cerebral palsy (CP) exhibit spasticity, muscle weakness, and immobility, in combination with an inability to deal with the effects of gravity. Thus, these children are at risk of developing muscle contractures, hip dislocation, windswept hip deformity (WS), and scoliosis, either in isolation or in combination.

Case Report: A 14 year old female patient started having pain over Right hip with gradual restriction of movements. Right hip was fixed in 30 degree of flexion, 20 degree adduction and 25 degree of internal rotation. Hip MRI was done which showed hyper intensity in superolateral portion of femoral head and corresponding part of acetabulum, ilio-psoas involvement. CRP was positive with ESR- 32mm/hr at the start of disease 1 year back. ASO titre <200, anti CCP 12.5 (equivocal), RA factor – negative, Hb- 9, TLC – 10,000. Broncho-alveolar lavage was done after which AFB smear, Cytology and Gene Expert was done and confirmed the diagnosis of Pulmonary TB with Rifampin sensitive. Montoux Test was done which was positive with 14 mm induration. Anti TB Cat I DOTS was started with bilateral surface traction. Now the patient is bearing weight with gradual recovery of painless joint motion of all the joint. She can now bear weight and can walk comfortably.

Conclusion: Poncet's disease is a diagnosis of exclusion, and differential diagnosis should be kept in mind; whenever joint involvement is seen with the presence of Tubercular foci elsewhere in the body.

Keywords: poncet's disease, tubercular rheumatism, juvenile idiopathic arthritis, rheumatoid arthritis, tubercular arthritis

Introduction

There has been much debate and diagnostic dilemma regarding diagnosis of a child which is not improving with anti-rheumatoid drug and clinical features are much related with Juvenile Idiopathic Arthritis. Poncet disease was first described in 1897 by Antonin Poncet ^[1], is a rare entity in which single or multiple joints are involved with features of arthritis. Very few cases have been described till now in literature and it does not find mention in standard text books. Most of the cases are from India, few from Brazil and China. Joint fluid analysis is sterile with no bacterial isolation and normal cell count.

Windswept pelvis is a rare feature seen in Poncet's Disease. This rare entity is found in cases with long standing disease. One hip is flexed, adducted and internal rotated while the contra lateral hip is flexed, abducted and external Rotated. Windswept hip deformity may occur in association with hip dislocation and scoliosis. Children with cerebral palsy (CP) exhibit spasticity, muscle weakness, and immobility, in combination with an inability to deal with the effects of gravity. Thus, these children are at risk of developing muscle contractures, hip dislocation, windswept hip deformity (WS), and scoliosis, either in isolation or in combination.

From a clinical perspective arthritis is the most common symptom of Poncet's disease, which can have a bilateral and symmetrical pattern. Other manifestations can present in the skin and also mucosa in the form of erythema nodosum, erythematous rash, papulonecrotic lesions, mouth ulcers and conjunctivitis.

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Case Report

A 14 year old female patient started having pain over Right hip with gradual restriction of movements. She was on NSAIDS since beginning of symptoms, but the disease process continued over time with subsequent involvement of Right knee, then Left knee in due course of 1 year. Joint was warm, tender with restricted movement and difficulty in weight bearing. She was put on prednisolone of 10 mg TDS for 1 months but symptoms still persisted. Diagnosis of Polyarticular Juvenile Arthritis was initially made and DMARD was started as the duration was more than 6 weeks and age was less than 16 years. Multiple symmetrical joints were involved by this time - both hip, both knee, both wrist joint and right ankle joint with sparing of small joints of hand and feet, axial skeleton and sacro-iliac joint was not involved. There was no history of loose stool, sore throat, UTI, contact with TB, chronic diarrhoea, heat intolerance. Morning stiffness was absent. However history of intermittent episode of fever was present. DMARD was continued for another 2 months but the symptoms continued to increase and by now the patient was totally crippled and bed ridden. There was no history of asthenia, fever, weight loss, cough, night sweats. Skin lesions were absent without any rashes, subcutaneous nodules, oedema, lymphadenopathy, jugular veins were not engorged Chest x-ray was advised and some ill defined lesion was present suggestive of consolidation in the right upper lobe. HRCT of chest was advised which further confirmed the diagnosis. HRCT Thorax was done which showed radio-opacity in left upper lobe with distal small nodular opacity. Similar opacity noted in right upper lobe. Rest of the lung field appears to be normal.

Patient was started with sulfasalazine, Hydroxychloroquine, NSAIDS. ANA, PT, INR was done and found to be normal. After 6 months Pt. developed asthenia, dry cough, fever and sinus tachycardia. Appetite decreased with rising ESR, so DMARD was stopped to evaluate for reactive arthritis secondary to TB. Nodular opacity was seen on chest X Ray. Broncho-alveolar lavage was done after which AFB smear, Cytology and Gene Expert was done and confirmed the diagnosis of Pulmonary TB with Rifampin sensitive. Montoux Test was done which was positive with 14 mm induration. Right hip was fixed in 30 degree of flexion, 20 degree adduction and 25 degree of internal rotation. Hip MRI was done which showed hyper intensity in superolateral portion of femoral head and corresponding part of acetabulum, ilio-psoas involvement. CRP was positive with ESR- 32mm/hr at the start of disease 1 year back. ASO titre <200, anti CCP 12.5 (equivocal), RA factor – negative, Hb- 9, TLC – 10,000. She developed gradual restriction of both hip joint movements. Windswept deformity developed in the pelvis. Right hip was 30 degree flexed, 25 degree adducted and 35 degree internal rotated with left hip 30 degree flexed, 35 degree abducted and 40 degree external rotated. Patient was totally bed ridden with inability to bear weight. Arthrotomy of hip joint was done with release of contracture around hip and synovial biopsy was done; report was non-specific synovitis with no granulomas and no features of rheumatoid seen. Anti TB Cat I DOTS was started with bilateral surface traction. Now the patient is bearing weight with gradual recovery of painless joint motion of all the joint. She can now bear weight and can walk comfortably.

Discussion

Tuberculosis is the most common prevalent disease in developing countries like India. Approximately 10% to 11%

of the extrapulmonary tuberculosis cases affect bones and joints, corresponding to 1% to 3% of all cases of tuberculosis. This entity is also called Tubercular Rheumatism, as tubercular infection is else where in the body, extra-articular and immune mediated inflammation mediated by HLA DR3 and DR4 auto-immune reaction. These patients have inherited defect in the chromosomal lineage and are at risk to autoimmune reaction. Several theories has put forward for this disease 1) Immunogenic reaction in which T cell mediated immune complex formation; settles down in different joints and starts inflammatory process. 2) Super-Antigen are formed which selectively causes articular cartilage destruction and sub-chondral lysis 3) Mycobacterial antigen have close mimic with articular cartilage proteoglycans 4) Hypersensitivity reaction to Mycobacterial antigen. This process is locally destructive but responds well to anti-tubercular drugs without any residual effect and recovers to normal function. Surgeon should have an eye to this diagnosis and should be kept in mind for treating unresolved cases. Poncet disease is actually a diagnosis of exclusion and is different from Tubercular Arthritis where there is no direct invasion of Mycobacterial infection and non-granulomatous inflammation.

Cases similar to ours have been reported in literature, where a diagnosis is made by exclusion, and is supported by the response to anti-tuberculous therapy, which delays the diagnosis of tuberculous rheumatism ‘‘Poncet’s disease’’ from 19 months to 12 years according to Rueda *et al.* [6] and Walter [7]. An important element in consolidating a diagnosis, is that by eliminating the focus of infection the pain and joint inflammation subside without leaving damage to the joints; this is borne out by this case. This element is part of the diagnostic criteria proposed by Rueda *et al.* [6] for Poncet’s disease, which include: (1) evidence of active extra-articular tuberculosis; (2) involvement of more than one joint, including knees and ankles; (3) no axial or vertebral compromise, or sacro-iliac involvement; (4) non-specific laboratory results; (5) complete remission after anti-tuberculous therapy; (6) no chronic joint compromise; and (7) the exclusion of other rheumatological diseases [6].

It is worth noting that, with modern diagnostic methods for tuberculosis infection, Poncet’s reactive arthritis might prove to be more prevalent than supposed. Currently, two diagnostic methods are used: CRP in sterile samples, such as synovial fluid; and interferon gamma release assays. Valleala *et al* [8]. have reported a case of tuberculosis diagnosed by use of that method, with confirmation through therapeutic response.

Conclusion

This case report is presented for the orthopaedic surgeon to keep in mind that arthritis of any large joint and windswept deformity of pelvis can mimic JIA in young child and diagnosis can be delayed due to ignorance. Due to high prevalence of Tuberculosis in developing countries like India Poncet’s disease should be kept in mind. About 20 percent of Tuberculosis is extrapulmonary and almost 2.5 million people worldwide die due to TB. There is wide variety of presentation to this disease. Even a latent infection elsewhere in the body can develop immunogenic reaction at the joints and develop pain, swelling and deformity. Surprisingly, these patients respond well to Anti-TB drugs with full recovery and normal function. Poncet’s disease is a diagnosis of exclusion, and differential diagnosis should be kept in mind; whenever joint involvement is seen with the presence of Tubercular foci elsewhere in the body.

Case Report



Fig 1: Xray of both wrist joint- Showing arthritic changes at wrist joint with decreased joint space



Fig 2: Xray of pelvis with both Hip joint showing windswept deformity of pelvis and arthritic changes with flexion, adduction and internal rotation of right hip and flexion, abduction and external rotation of left hip



Fig 2: Xray of both Knee joint showing early degenerative changes Grade I

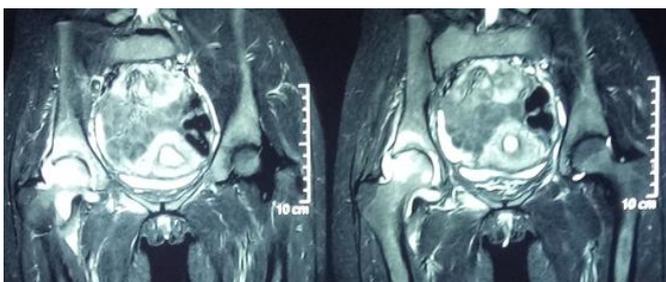


Fig 3: MRI of pelvis showing degenerative changes in supero-lateral portion of femoral head and acetabulum.

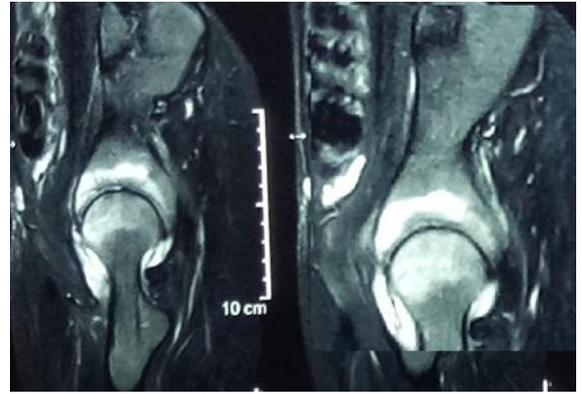


Fig 4: MRI of Pelvis showing superior part of head involvement and corresponding acetabular involvement with diminished joint space

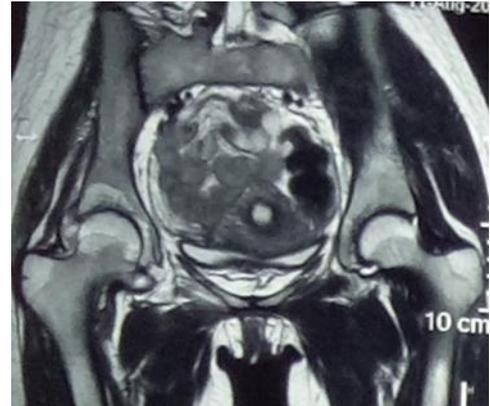


Fig 5: MRI of Pelvis with both Hip Joint



Fig 6: CECT Thorax showing Non-enhancing lesion seen in posterior segment of right upper lobe and apicoposterior segment of left upper lobe



Fig 7: X Ray of Chest is within normal limit

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