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Pseudo-Septic Hip Arthritis as the Presenting Feature of Juvenile Ankylosing Spondylitis: A Case Report

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ABSTRACT

Background: *Juvenile onset AS differs from the adult form of the illness in that peripheral joint (like hip) arthritis is much more common and can precede, by many years, the onset of inflammatory back disease.*

Case characteristics: *A young male patient of 15 years started having hip pain (Right>Left) associated with limping. Initially he was diagnosed as a case of right side TB hip but anti tubercular therapy failed to improve his symptoms. After about 3 years he was again diagnosed as bacterial septic arthritis, this time of the left hip and managed for that. Finally the diagnosis of Juvenile Onset Ankylosing Spondylitis (AS) was made based on modified New York criteria.*

Intervention: *Emphasis was placed on the importance of long term home exercise program.*

Outcome: *At a 2-month follow-up visit, the patient gained significantly in his ease of performing activities of daily living. Also he was referred for surgical spinal osteotomy for kyphosis correction and/or bilateral total hip replacement.*

Message: *Pseudo septic hip arthritis as the presenting features of AS has been quite rare but may be expected in the juvenile onset variant.*

Abbreviation: *AS: Ankylosing spondylitis , B/L: Bilateral , TB: Tuberculosis , ATD: Anti-tubercular drugs ,CRP: C Reactive Protein ,ESR: Erythrocyte Sedimentation Rate, HLA: Human Leucocyte Antigen ,TNF: Tumor Necrosis Factor ,ROM: Range of motion ,SI Joint: Sacro-iliac Joint, MRI: Magnetic Resonance Imaging , DL Spine: Dorso-lumbar Spine*

KEYWORDS : Juvenile onset Ankylosing Spondylitis, Pseudo septic hip arthritis.

Introduction:

Ankylosing spondylitis (AS) is a seronegativespondyloarthropathy characterized by axial arthritis, ocular inflammation, enthesitis, and peripheral oligoarthritis.[1]Juvenile ankylosing spondylitisdiffers from the adult form of the illness in that peripheral joint (hip, knee, ankle or even shoulder)arthritisorenthesitis is much more common and can precede, by many years, the onset of inflammatory back disease.[2] In addition, functional status may be better preserved in juvenile onset as opposed to adult-onset cases.[3]However, in contrast to the classical changes of AS in the spine, subchondral bone marrow inflammation in the hip joint of Juvenile AS does not lead to formation of new bone, but results in an erosive disease, which will often destroy the joint.[4] Although the hips are frequently involved during the course of AS, initial presentation with severe hip arthritis is unusual, and abrupt-onset hip arthritis mimicking septic arthritis in AS has been quite rare.[5]Here, we report a case with Juvenile AS who presented initially as pseudo-septic hip arthritis.

Case report:

About 6 years ago in 2009, the then 15 year old male patient (Figure 1) started having bilateral (B/L) upper thigh pain, B/L hip pain (Right > Left), LBP (mostly left lumbosacral and sacroiliac region) and inter-scapular spinal pain. The pain was more in the morning and improved over the day.Morning stiffness lasted for about 2 hours. He gradually faced difficulty in walking, cycling, stair climbing and developed a right sided limp. Important negative history includes no history of (H/O) awakening at night due to pain, no H/O eye, skin, nail, hair, gastrointestinal or genital involvement, no H/O small joint pain or swelling, no H/O bladder or bowel involvement and no H/O similar involvement among any family members. He had H/O occasional left heel cord pain, alternate buttock pain and B/L anterior chest wall (more near chondro-sternal joint) pain over the last 5 years. Also had arecent episode of urinary burning sensation.

He was initially diagnosed by doctors at his hometown to haveTuberculosis (TB) of right hip (a type of septic arthritis) on the basis of high ESR (33 mm at 1st hour and 65 mm at 2nd hour) and borderline positive IgM and IgG Antibody to Mycobacterium species. X- ray done on February of 2010 showed joint space diminished in right hip associated with marginal sclerosis (Figure 2).But some points were against TB also, viz. Manteaux test and CRP were negative as well as sputum examinations on two occasions (April and May 2010) were negative. The pelvis X- ray previously mentioned also show joint space narrowing of contralateral left hip. X ray DL spine was reported as normal but on close scrutiny show features like vertebral squaring, marginal sclerosis and osteopenia (Figure 3).Eventually he was started with 4 drug chemotherapy and leg traction with 6 pound weight on the basis of diagnosis of TB right hip. Patient took the traction for 2 days and Anti-tubercular drugs (ATDs) for 4 months in 2010 but his symptoms were showing no signs of improvement.

So in 2011 he started taking some expensive Ayurvedic medicines and continued it for 9 months. Apparently he had an improvement in his walking speed and could cycle also. But the benefit was not sustained and he very quickly returned to his previous status.

In 2012 when the patient came to a superspeciality Hospital at Kolkata, his initial reports suggested a quiescent disease status (ESR and CRP normal). But after few months, both acute phase reactants were found to be raised and it remained so throughout the year. Also patient had resorted to stick for walking and gradually showing loss of mobility of B/L hip as well as spine and chest. Other investigation findings included: HLA B27 negative, Manteaux test borderline positive, peripheral blood film hypochromic microcytic anisocytosis with hemoglobin level borderline normal. X rays showed hip joint space reduction (R>L), irregular acetabular margin, enostomosis of right hip bone, enthesopathy of B/L trochanters and non-visualization of SI joints (Figure 4). This time he was diagnosed as juvenile onset ankylosing spondylitis based on modified New York criteria but biologics could not be initiated due to financial constraint.[6] He was rather put on regular anti-inflammatory medications, ROM exercises and mandatory daily prone lying.

But, probably due to non-initiation of TNF inhibitor medication, his B/L hip inflammation continued to mimic picture of septic arthritis. MRI of pelvis was done in 2013 due to increased left hip painwhich showed reduced joint space and sclerosis of both hip joints (R>L) along with patchy signal intensity and synovial enhancement of left hip suggestive of infective etiology. So, this time there was an impression of pseudo septic arthritis ofleft hip. Also there wereincidental findings of multilevel disc bulge with neural foraminal narrowing at L3-4 and L4-5 level. Minimum canal diameter was 12.1 mm at L4-5.

There was no comment on SI joint (Figure 5). So, beside a brief period of bed rest, a course of systemic broad spectrum antibiotic was also initiated.

In the spring of 2014, his investigations were repeated in our Institute. Significant findings include HLA-B27 positive (compared from negative 1 year back), hemoglobin 11.8 gm% and ESR 58 mm/hour. X ray pelvis showed Grade 3 sacroilitis on right side and Grade 4 sacroilitis on left side (Figure 6). X ray DL spine showed squaring of vertebrae, straightening of spine, marginal sclerosis but no syndesmophytes (Figure 7). X ray chest appeared to be within normal limits (Figure 8). Clinical symptoms included LBP of inflammatory nature, left hip pain, inter-scapular spinal pain and alternate buttock pain. On inspection there is forward craning of the neck, increased dorsal kyphosis, rounding of the shoulders, decreased lumber lordosis, wasting of the buttocks and flattening of the chest. On palpation no tender or swollen peripheral joint noted, including the 44 joint ASAS core set, manubri-sternal joint, chondro-sternal joint, Greater Trochanter, IschialTuberosity, TibialTuberosity, Iliac crest or Heel cord.[7]However tenderness was present over left SI joint. Pelvic compression test was also positive on left side.Also except a small amount of internal rotation and external rotation (about 10 ° of both), all other hip movements of both hip were restricted. The different spinal mobility parameters are shown in the Table 1.

Table 1: Spinal mobility

Serial number	Tests	Value in patient	Normal range
1	Occiput to wall distance (Flesche test)*	8 cm	0 cm
2	Tragus to wall distance*	19 cm	
3	Cervical rotation (right, left and mean)*	50°and 70° respectively i.e., mean 60°	70-90°
4	Chest expansion*	2 cm	>5cm
5	Macrae and Wright modification of Schober's index*	4 cm	>5cm
6	Finger to floor distance*	28 cm	
7	Lumbar lateral flexion (mean of right and left)*	16 cm	>10 cm
8	Intermalleolar distance*	11 cm	

*[*Definitions: 1) Occiput to wall distance (Flesche test): Horizontal distance between occiput and wall, patient standing with heels and buttocks against the wall. 2) Tragus to wall distance: Horizontal distance between right tragus and wall, patient standing with heels and buttocks against the wall without rotation. 3) Cervical rotation: Distance between tip of nose and acromioclavicular joint in neutral less the same distance in maximal ipsilateral rotation. The mean of right and left results being calculated. 4) Chest expansion: The difference in centimetres to the nearest 0.1 cm between full expiration and full inspiration, measured at the nipples. 5)Macrae and Wright modification of Schober's index: The midpoint of the posterior superior iliac spines roughly correspond with dimple of venus. Further marks are placed 5 cm below and 10 cm above this lumbosacral junction. The patient is asked to bend forwards as far as possible, keeping the knees straight, and the distraction between these 2 marks is recorded. 6) Finger to floor distance: Distance between tip of middle finger and the floor following maximal lumbar forward flexion with knees extended. 7) Lumbar lateral flexion: Distance between tip of ipsilateral middle finger and the floor following maximal lumbar lateral flexion, with both feet on the floor, knees extended and without rotation The mean of right and left results being calculated. 8) Intermalleolar distance: Patient supine, the knees straight and the feet pointing straight up. The patient is asked to separate the legs as far as possible and the distance between the medial malleoli is measured.*]

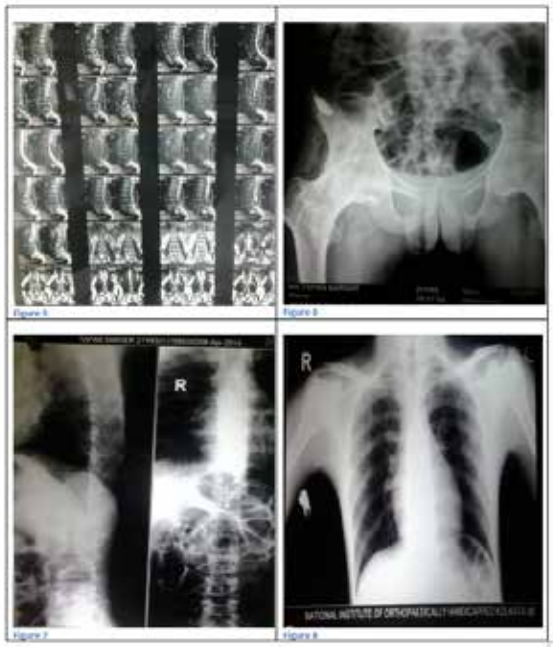
So, again the diagnosis was of juvenile onset ankylosing spondylitis. In our Institute, he was provided with a home program of trunk mobility exercises, stretching, lower and upper extremity strengthening. Emphasis was placed on the importance of prone lying, swimming and long term habit of exercise. At a 2-month follow-up visit, the patient reported he was pain free and gained significantly in his ease of performing activities of daily living. Also he was referred for surgical spinal osteotomy for kyphosis correction and/or bilateral total hip replacement among which the former operation generally precedes the later in such cases.[8]

Discussion:

Juvenile ankylosing spondylitis, defined as onset of disease prior to age 16, is considered a pediatric form of ankylosing spondylitis also known as HLA-B27-related arthritis, Marie-Strumpell's disease, von Bechterew's disease orpelvospondylitisossificans.[9]Bowyer reported that juvenile patients were seen by an average of 2.5 physicians before being correctly diagnosed with juvenile ankylosing spondylitis.[10]The reason behind that is frequent early onset of peripheral joint involvement and delayed axial changes that may not occur until years later in case of juvenile AS.[11] Hence a vigilant approach is necessary for early diagnosis or unless the young patient can develop advanced joint destruction.

Prior reports of pseudoseptic hips as presenting feature of juvenile AS is almost non-existent. T. Mathur et al evaluated the charts of all patients who were diagnosed with AS in the rheumatology clinics at John H StrogerJr. Hospital of Cook County (Chicago, IL) from the years 2006 to August 2009. They, found that 10% (11 of 110) of AS patients presented with severe hip involvement and three of those presented with symptoms of acute-onset hip pain with clinical and laboratory features that mimicked bacterial/tubercular arthritis, so-called "pseudoseptic arthritis". But unlike those 3 cases, our patient has been twice diagnosed as septic arthritis (first time TB hip and the next time bacterial septic arthritis of hip).[5]

Our patient also developed significant B/L hip joint destruction probably due to delayed diagnosis and consequent inability to start injectable anti–TNF-α agents or sustained anti-inflammatory medication as well as proper physiotherapy. Total hip replacement is the most common surgery performed in patients with ankylosing spondylitis and our patient will also need that. [12]But he may need spinal osteotomy for kyphosis correction before hip replacement as suggested by G Q Zhenget al.[8] Also, revision surgery may be necessary, because the patient is very young and quite active.



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