Rapidly destructive coxarthrosis as early presentation of Rheumatoid Arthritis in a young women: A case report.

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Abstract

Rapidly destructive coxarthrosis of the hip joint is a rare condition of which the etiology and pathogenesis are poorly understood. Ordinary arthrosis or osteoarthritis of the hip joint usually occurs over long duration with well-established risk and predisposing factor in the elderly. However, rapid destruction may occurs over a period of a few months' damaging the acetabulum and femoral head. Various mechanism has been reported in previous literatures which implicates both subchondral insufficiency and autoimmune related. We report a 36-year old Indian woman with progressive left hip pain as early symptoms of rheumatoid arthritis with negative rheumatoid factor and positive anti-citrullinated peptide antibody, supported by the magnetic resonance imaging and synovial histopathological findings. There was only one similar case reported in the literature and more than eight cases with pre-existing rheumatoid arthritis occurring in patients of more than 50 years of age.

Keywords: Rapidly destructive coxarthrosis, Rheumatoid Arthritis, Magnetic resonance imaging, Histopathology.

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Introduction

Arthrosis is synonymous to osteoarthritis (OA) which is common degenerative disease of a joint in elderly and coxarthrosis specifically refer to hip joint osteoarthritis and it can be primary or secondary of which the latter due to other inflammatory joint disorders. Both arthrosis and arthritis shared some common clinical features such as pain and tenderness, stiffness and limited range of joint motion although radiologically may differentiate these conditions.

Rapidly destructive coxarthrosis (RDC) was first described by Postel and Kerboull [1] as a rare condition commonly affecting the hip joint in the elderly. Though the etiology and pathogenesis are yet to be elucidated, it is usually unilateral and destructive within a short period of time from the onset of symptoms.

Diagnosis is usually made by exclusion as there are no diagnostic markers identified. There are several inflammatory joint complaints including degenerative disorders that have been implicated in the development of this condition. Eight cases have been reported with RDC with underlying pre-existing RA of more than two years duration and one case reported as an initial manifestation of seronegative RA which fulfilled the RA classification criteria. All the patients reported were more than 45 years of age.

Our case report describes a young women who presented with only RDC of the left hip joint as an early presentation of negative rheumatoid factor and positive anti citrulinated peptide antibody rheumatoid arthritis.

Case Presentation

A 36-year old Indian women, an engineer by profession, presented with history of progressive left hip pain which was very disabling for over 2 months' period. The onset of pain began 4 years ago after her second delivery. She was having difficulty in ambulation due to the unbearable pain which was localized to the left hip and aggravated by movement. Pain was relieved with rest and temporarily resolved with non-steroidal anti-inflammatory drugs (NSAID). She was not on any corticosteroid prior to the ailment and had no history of injury. She had no arthritis over other peripheral joints present nor has she had any other systemic manifestation of connective tissue disease. She was then given prednisolone 5 mg daily for 2 months duration without any significant improvement.

Laboratory investigations showed raised erythrocyte sedimentation rate (ESR) (66 mm/hour), highly sensitive C-reactive protein (hsCRP) (8.93 mg/L), but negative rheumatoid factor (RF) and normal hematologic panel. Synovial biopsy taken from the affected hip 4 years after magnetic resonance imaging (MRI), were consistent with severe chronic synovitis changes (Figure 1). Culture and sensitivity including for *Mycobacterium tuberculosis* were negative.

Plain x-ray of the hip joints showed narrowed joint space with presence of effusion and erosion of the femoral head and acetabulum left hip (Figure 2). The right hip joint was normal. MRI of the left hip joint supported the presence of effusion in more details (Figure 3). Plain x-ray of both hands was normal.

Based on the histopathological findings, she was diagnosed with seronegative rheumatoid arthritis (RA) and given methotrexate with indefinite duration but she defaulted treatment since her symptoms had remained the same. Subsequently she was referred to a rheumatologist for further management. Her left hip was still in pain, severe enough to restrict her daily activities. However, her other joints remained normal. Further diagnostic procedure showed raised anti citrulinated peptide (anti-CCP) of 187.3 IU/L (normal <0.5 IU/L).

Methotrexate was recommenced at 10 mg weekly. Although biologic disease modifying agent (bDMARD) was suggested, the patient refused. Over the ensuing two month's period, she showed marked improvement without untoward effects

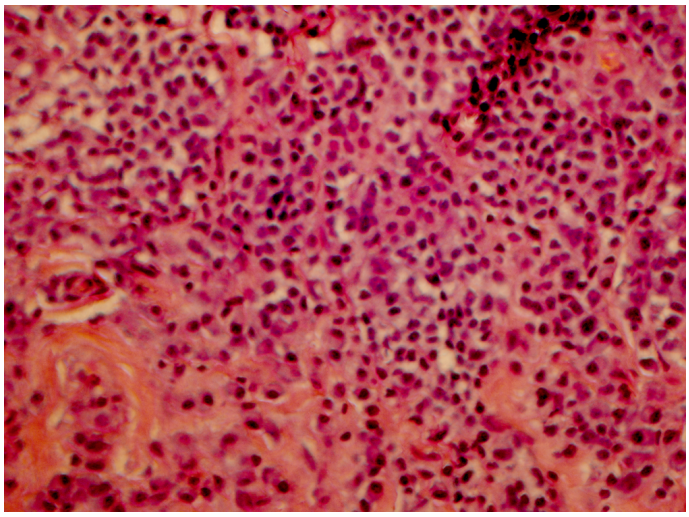

Figure 1. *Synovial tissue from left hip joint showing marked vascular congestion and very dense inflammatory infiltrate. There was no evidence of villous pattern or haemosiderin pigment formation. The synovium lined by reactive histiocytes and a stroma densely packed with plasma cells and matured lymphocytes. Neutrophils, eosinophils, and malignancy cells were not seen.*

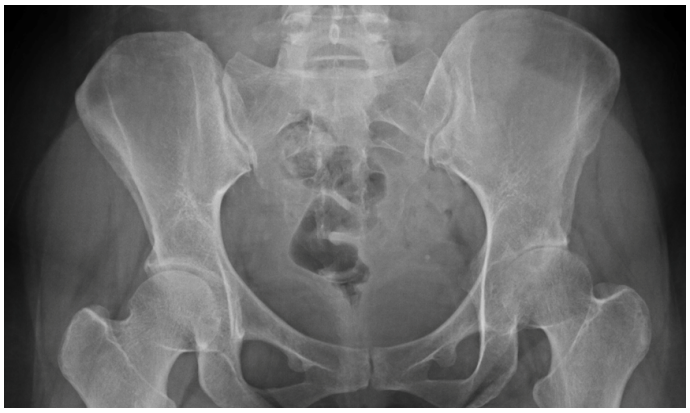

Figure 2. *Plain radiograph of the pelvis showing reduced left hip joint space with lucent areas within the left femoral head and in the adjacent acetabulum (Arrow).*

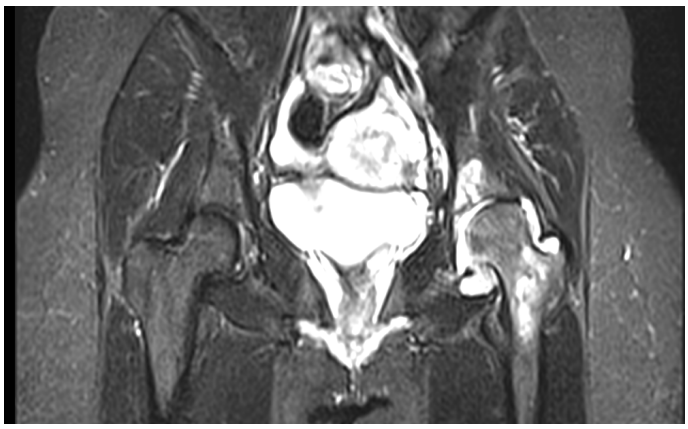

Figure 3. *Contrast enhanced coronal image showing abnormal enhancement in the left acetabulum, femoral head and greater trochanter and within the hip joint effusion (Arrow).*

of the methotrexate. Her pain and health assessment scores were almost 0. She opted for medical treatment despite being recommended for reconstructive surgery. She has remained in remission since then.

Discussion

RDC of the hip joint has been described as a rare condition compared to ordinary osteoarthritis. The actual etiology and pathogenesis of the rapidity of the destruction is still illusive [2]. Hence, early identification of this condition often failed and usually total hip arthroplasty will be performed at early stage. Previous literatures hypothesized presence of subchondral insufficiency fracture may explained the rapid destruction [3-8]. However, in recent studies, immune mechanism has been implicated in the pathogenesis of osteoarthritis. An inflammatory synovium/synovitis with T- and B-cells and macrophages infiltrates, presence of pro-inflammatory cytokines, immunoglobulins, immune complexes, complement activations has been linked to increased cartilage damage and pain in large number of OA patients [9-15]. Inflammatory joint disorders such as RA have been reported in a few RDC cases [16-20]. Most of these cases are elderly patients (age >45 years), with established RA of more than two years. Only 1 case (age >50 years) which was described as an initial presentation of RA but with other joint involvement as well which fulfilled both 1987 and 2010 American College of Rheumatology (ACR) and European League of Rheumatism (EULAR) classification for RA [21,22]. However, the RF and anti CCP were both negative in the latter case. Anti CCP antibody has not been described in degenerative joint disease in previous literature and it is more sensitive and specific with prognostic value in RA [23].

Our case, is the first case with RDC of the hip joint as initial manifestation of RA at a very young age (32-years at onset of pain) without any other joint or systemic involvement. However, this case did not fulfill the ACR/EULAR classification criteria for RA except for positive anti CCP antibody. Although osteoarthritis had been entertained as the primary diagnosis earlier based on the radiological features, the characteristic i.e. the rapidity and severity of the destructive features and demography (young age, not overweight, unilateral involvement, without trauma) is not in favor of OA per se. Histopathological and MRI findings with additional anti CCP positivity despite negative RF narrowed it down to RA. She may be considered fortunate that despite the delayed diagnosis of RA (4 years after onset) and inadequate response to methotrexate due to non-compliance, she did not have any systemic involvement or peripheral arthritis over the years.

It has to be admitted that MRI has become a very useful supportive diagnostic tool in RDC. MRI of the hip in RDC has been well described by Boutry et al. who have focused on the main key signs i.e. joint effusion, bone marrow edema like in the femoral head and neck or acetabulum or both, femoral head flattening, and cyst like subchondral defects [24]. These findings are similar to that of our patient's MRI. However, there are no features suggestive of bursitis in this patient as reported in previous cases [25].

We have thus described the first case of RDC in a 36-year old women with an early manifestation of seronegative RA with positive anti CCP, and without other joint or systemic involvement who responded well to methotrexate at 10 mg weekly dose.

Based on this case report, we conclude that it is imperative to diagnose RDC as early as possible in RA, non-RA as well

as other inflammatory conditions in patients presenting with unilateral hip pain which is unique and rare as it could lead to poor outcome if left untreated at the early stages. Reconstructive surgery or arthroplasty, hence, could be deferred.

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