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Co-existing Ankylosing Spondylitis and Rheumatoid Arthritis in a Patient Undergoing Total Knee Arthroplasty Under Peripheral Nerve Blocks

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Learning Point of the Article:

The coexistence of AS and RA is rare; and clinicians must remain suspicious of this entity while following up patients with either diagnosis, as a diagnosis of one does not rule out the other, and also influences treatment choices.

Abstract

Introduction: While laboratory tests and clinical criteria are well-defined, simultaneous presence of concomitant symptoms can lead to a misdiagnosis and delay in initiation of appropriate pharmacotherapy. Similarly, natural joint disease progression may also vary and present with atypical presentations.

Case Report: Here we report a case of Ankylosing Spondylitis (AS) with Rheumatoid Arthritis (RA) proven clinically, radiologically, as well as serologically, in whom multiple joint replacement procedures were performed. The association between these two conditions with overlapping features and laboratory markers constitutes an interesting phenomenon and can pose a diagnostic dilemma, thus, increasing the importance of awareness and early diagnosis of this co-existence. We report the case of a 63 year-old gentleman who was a known case of ankylosing spondylitis since the age of 38. The patient had undergone a staged bilateral total hip arthroplasty 18 years ago and presented to use with knee arthritis necessitating Total Knee Arthroplasty. The patient was subsequently diagnosed to have coexisting Rheumatoid Arthritis. After a failed attempt for spinal anaesthesia, a para-sacral sciatic block in the lateral position and an infra-inguinal femoral nerve block and obturator nerve block in the supine position were given under ultrasound guidance. The total knee replacement was performed successfully without any intra-operative complications.

Conclusion: This combined entity requires accurate assessment or can be easily misleading. No definite conclusion is possible with regards to the etiopathogenesis of these conditions coexisting but this association with overlapping features and laboratory markers constitutes an interesting phenomenon and can pose a diagnostic dilemma.

Keywords: Ankylosing spondylitis, rheumatoid arthritis, arthroplasty, hip, knee.

Introduction

Before the identification of the rheumatoid factor, Ankylosing Spondylitis (AS) was considered an axial or spinal variant of Rheumatoid Arthritis (RA) and was a part of the RA spectrum, termed as "rheumatoid spondylitis".[1,2].

Overlapping features made the differentiation of these two conditions difficult. Several features have been established for distinguishing these two conditions with each having its criteria such as the modified New York criteria for diagnosis of AS (1984), Assessment in Ankylosing Spondylitis (ASAS) group criteria for axial spondyloarthritis (2009) and RA classification criteria of the ACR/EULAR (American College of Rheumatology/ European League Against Rheumatism) of

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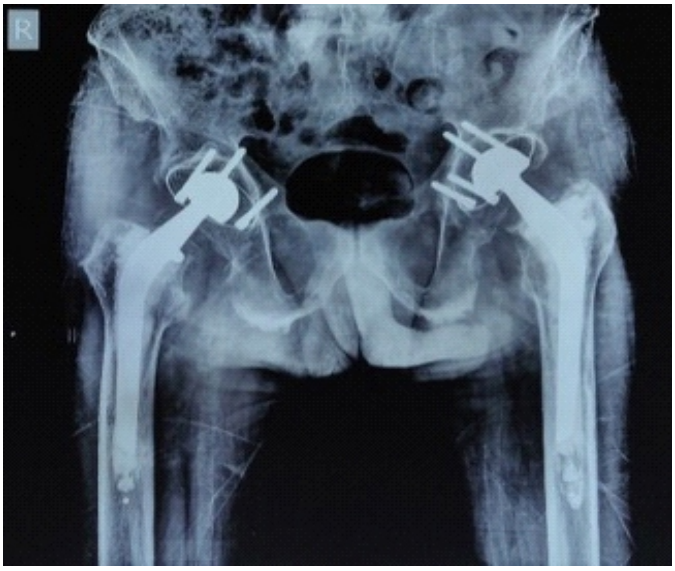

Figure 1: Radiograph of the pelvis shows bilateral total hip arthroplasty with prostheses in situ.

2010. The probability of a person developing both AS and RA was estimated to be 1 in 1,00,000 [1]. Neither does accidentally detected HLA-B27 in patients with RA increase the incidence of sacroiliitis nor does the presence of Rheumatoid factor in patients with AS increase the risk of peripheral arthritis.[3]

Case Report

A 63-year-old gentleman presented to our institution with chief complaints of bilateral knee pain (left more than right) which was insidious in onset and gradually progressive over the past 2 years. The patient had undergone staged bilateral total hip arthroplasty 18 years ago at our institution (Fig. 1). The patient was a known case of ankylosing spondylitis which was diagnosed when he was 38 years old. He also complained of back pain, pain in multiple small joints of hands and feet along with generalized stiffness which exhibited diurnal variation, being more in the morning and less at night. The pain was maximal upon initiating movement after a period of rest and would get better with movement. Family history was not significant.

His cervical spine and lumbar spine were stiff. Chest expansion was less than 2 cm. There was an ulnar deviation of both wrist joints with hyperflexion of the metacarpophalangeal joints and hyperextension of the proximal and distal interphalangeal joints. There were no Bouchard or Heberden nodules (Fig. 2a). Joint line tenderness was present on both knees. The left knee joint had a fixed flexion deformity of 30º with further flexion up to 90º. Schober's test was positive.

HLA B27 and Rheumatoid factor were positive. Anti-CCP antibodies were negative. ESR was 103 mm/hour. CRP was 80.6 mg/L.

Radiographs of the Pelvis with both hips showed complete fusion of bilateral sacroiliac joints. Both hip prostheses showed no evidence of loosening or subsidence. Both wrists and hands showed diffuse osteoporosis, ulnar subluxation, and destruction of the carpus with severe erosions and mutilation of the distal ulna and radius. There was subluxation of the metacarpophalangeal joints along with symmetric erosion, destructive changes, and joint space narrowing of the metacarpophalangeal, proximal and distal interphalangeal joints as well (Fig. 2b). The lumbar spine revealed squaring of vertebrae, vertebral scalloping, and fusion of the vertebrae as evidenced by the ossification of the annulus fibrosis and anterior longitudinal ligament (Fig. 3a). The cervical spine

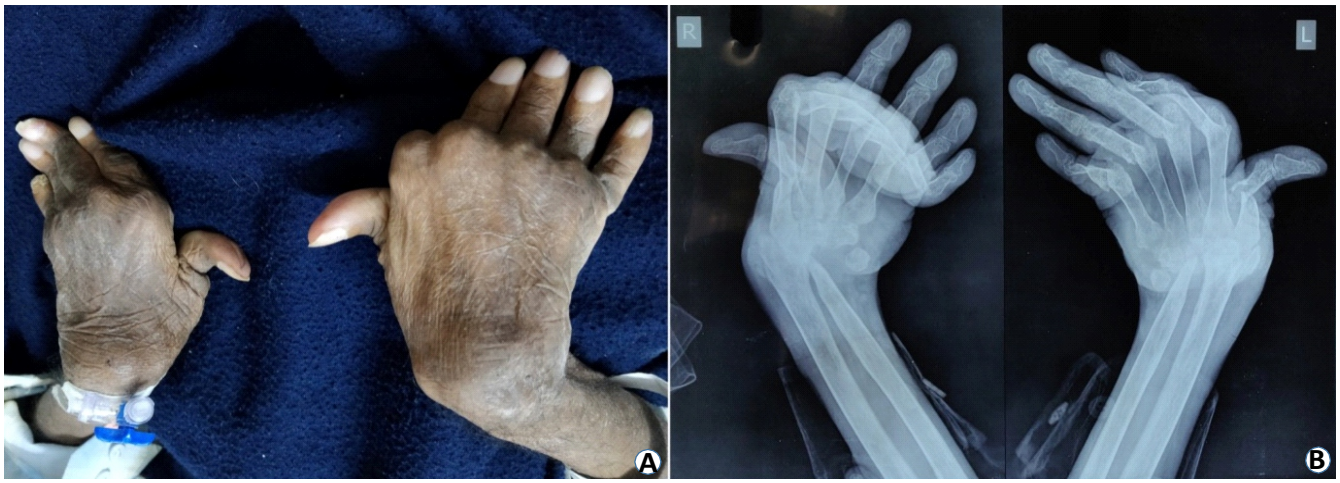

Figure 2: (A) Clinical picture shows ulnar deviation of both wrist joints with hyperflexion of the metacarpophalangeal joints and hyperextension of the proximal and distal interphalangeal joints. (B) Radiograph of both hands showing diffuse osteoporosis, ulnar subluxation, and other destructive changes of the carpus.

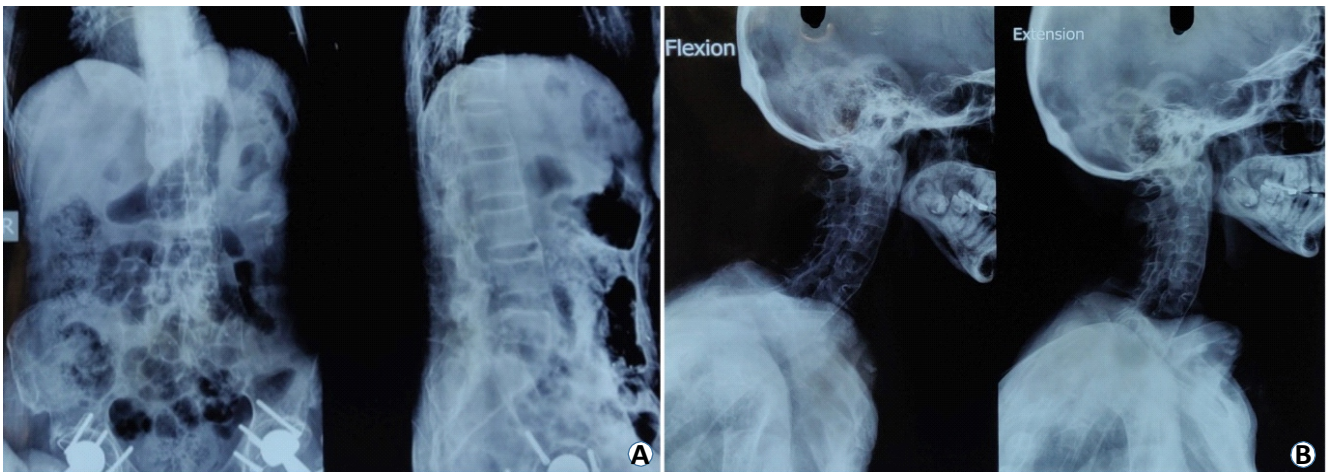

Figure 3: (A) Radiograph of the lumbar spine showing squaring of vertebrae, vertebral scalloping, and fusion. (B) Radiograph of the cervical spine showing ossification of the anterior and posterior elements along with ossification of the disc space.

revealed complete ossification of the anterior and posterior elements along with ossification of the disc space (Fig. 3b). Knee radiographs showed concentric joint space reduction with peri-articular osteopenia (Fig. 4a,b). At presentation, the patient was wheelchair-bound. Till 1 year ago, the patient was a community ambulator. Thereafter, there was a progressive decline in his ambulatory status.

Due to the unusual involvement of the hands and wrist, we performed additional investigations. His ACR/EULAR RA classification criteria (2010) score was 7, which indicated a diagnosis of Rheumatoid Arthritis. As per the modified New York criteria, our patient fulfilled the clinical and radiological criteria for a diagnosis of AS. In addition, our patient also fulfilled the criteria for a diagnosis of AS according to the ASAS classification criteria for axial spondyloarthritis. Thus, we diagnosed our patient as having coexisting ankylosing spondylitis and rheumatoid arthritis.

After evaluation by our physician and anesthetist, the patient was planned for a Left total knee arthroplasty. Before surgery, since neck movement was absent and mouth opening was 1 cm, a difficult airway was anticipated and hence a provision for fibre optic intubation was arranged. In view of the poor pulmonary function tests [forced vital capacity (FVC) - 37%, forced expiratory volume in 1 second (FEV1) - 50%, FEV1/FVC - 1.35], the high risk of post-operative ventilation was explained to the patient and relatives, and consent was taken for the same. Due to the poor respiratory function and difficult airway anticipated, it was decided to attempt spinal anaesthesia, failing which a sciatic and femoral nerve block would be given; and general anaesthesia was planned as the last option, even with fibreoptic intubation kept available. A trial of spinal anesthesia was given in the lateral position as the sitting position was

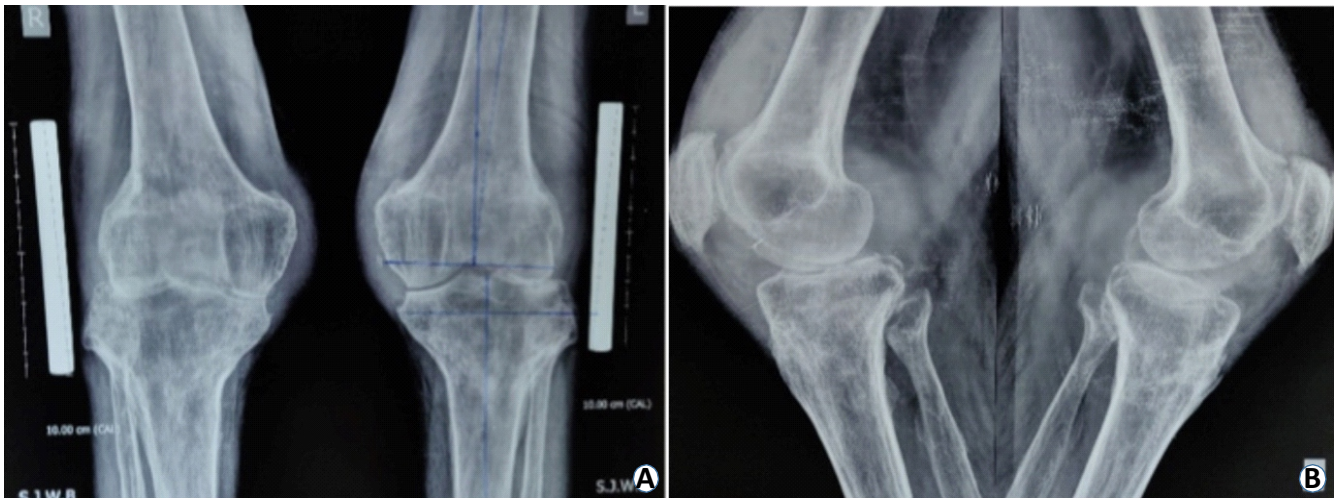

Figure 4: Radiographs of the knee showing concentric joint space reduction with peri-articular osteopenia in anteroposterior (A) and lateral (B) views.

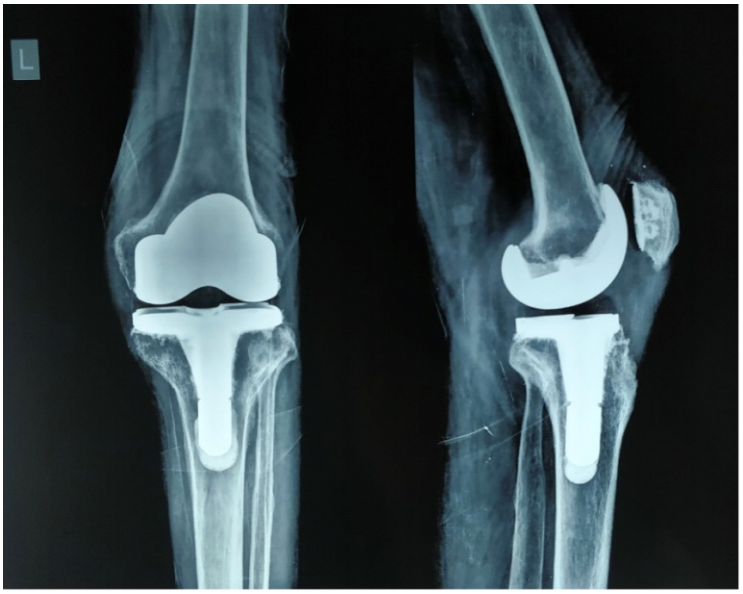

Figure 5: Post-operative radiograph of the left knee after the total knee arthroplasty procedure.

difficult due to structural deformities. However, it could not be performed successfully as the ligaments were ossified. Next, a para-sacral sciatic nerve block (Ropivacaine 0.5% - 12 mL and Clonidine 15 micrograms) in the lateral position, and an infra-inguinal femoral nerve block (Sensoricaine 0.375% - 30 mL + Clonidine 30 microgram) in the supine position were given under ultrasound guidance. The patient was sedated with Inj. Midazolam 1 mg and Inj. Ketamine 20 mg given in small aliquots, to prevent movement of the contralateral lower limb. The total knee arthroplasty was performed successfully without any intra-operative complications, only under peripheral nerve blocks. (Fig. 5). Post-operatively the patient was made to walk full weight bearing with walking frame support and was started on Sulfasalazine 500 mg once a day.

Discussion

The probability of both AS and RA coexisting in the same patient is about 1 in 1,00,000. Though cases with the concurrent presence of both conditions have been described, most of these were described more than 30-40 years ago when there was a lack of specific laboratory tests and diagnostic criteria.[1] Azevedo published the first case report using the updated Assessment in Ankylosing Spondylitis (ASAS) group criteria for axial AS and the 2010 American College of Rheumatology/ European League Against Rheumatism (ACR/EULAR) criteria along with anti-CCP testing for RA, which had not been done in previous studies.[1]

Features distinguishing these 2 conditions: Age of onset for AS is 20-45 years as compared to 40-70 years for RA; AS is associated with HLA-B27 gene while RA is associated with HLA-DR4 or DR1 genes; AS has a male preponderance while RA has a female preponderance; AS has a predilection for axial joints mainly the lumbar spine and sacroiliac joints, large joints (hip, knee, shoulder) and distal interphalangeal joints with asymmetric involvement whereas RA affects the peripheral small joints of the hands and feet (except DIP) in a symmetrical fashion with occasional involvement of the cervical spine (atlantoaxial subluxation).[4]

As per Azevedo et al, most of the previously published reports only used clinical data and lacked laboratory tests, with theirs being the only case using the latest criteria in addition to anti-CCP testing which was not done in any previous reports.[1] Extra-articular manifestations of either disease can also help in the additional diagnosis and further management.[5] The presence of an extra-articular manifestation of AS in a patient with RA or vice versa warrants further laboratory testing.[6] Since there is considerable overlap in the signs and symptoms of AS and RA, one should be skeptical in diagnosing both diseases together. The age of presentation can again be a confounding factor. Surgical implications in such patients should also be kept in mind when considering such patients for general anesthesia as also for post-operative recovery to avoid prolonged immobilization and an appropriate rehabilitation program.[7]

Fallet et al considered this coexistence to be unlikely and that this would warrant revisiting the basic concepts that define these two conditions.[8] Another theory is that both diseases may often co-exist, but the initiation of drug therapy on one of them manifesting suppresses the second disease. The second disease may manifest earlier if the patient discontinues treatment or on clinical evaluation of a long-term follow-up which was also seen in our case. This highlights the need for periodic follow-ups of patients presenting with a mixed picture which may warrant alteration in the drug regimen as well.[2] TNF inhibitors are efficacious in both diseases. However, in RA, disease-modifying antirheumatic drugs (DMARDs) are initiated at the earliest, and these drugs cannot be prescribed in AS in the absence of peripheral joint involvement.[9] Awareness and early diagnosis of the coexistence of these two diseases is also important therapeutically as agents such as gold and chloroquine are known to be beneficial in RA but are not helpful in AS.[10] This perhaps warrants the need for devising distinct therapeutic regimens for such patients with coexisting AS and RA.[11] In vitro, the Epstein-Barr virus has been shown to stimulate B lymphocytes in AS patients to produce the rheumatoid factor.[12] Taking into account these findings it could be possible that certain inhibitory mechanisms prevent the occurrence of a positive RF in AS patients while also being

present purely by chance.[3] Thus, it cannot be ascertained whether these two diseases coexist coincidentally or whether there is a genetic predisposition. This could also perhaps explain why every patient with HLA-B27 and HLA-DR4 positivity does not develop AS and RA respectively.

In clinical practice, difficulties do arise as up to 50% of patients with AS can have peripheral arthritis with relative sparing of spinal involvement, and also up to 30% of patients with RA can have SI joint involvement. This raises concerns with regards to the extra-articular manifestations, treatment, and complications which differ between these 2 groups. This is precisely why the recognition of the association between HLA-B27 and AS was a significant landmark.[8] Amongst patients with RA, about 9% have been proven to be HLA-B27 positive. However, this does not predispose this set of patients to develop sacroiliitis any more than the population which is purely HLA-B27 positive.[8] Since 50% of patients with AS can have peripheral joint arthritis and patients with RA can also develop ankylosis of cervical and SI joints (20%) in the advanced stage, it can become difficult to distinguish these cases. This is where laboratory testing for rheumatoid factor, anti-CCP, and HLA-B27 become extremely invaluable.[10] The presence of rheumatoid nodules is a clinical clue to clinching the diagnosis since rheumatoid nodules are rarely seen in AS.[13] Similarly, another clue for picking up RA when peripheral arthritis is seen in AS is the symmetrical involvement and erosive pattern. In addition, sacroiliitis though seen in RA is rarely the initial presentation, and even rarer is the occurrence of lumbar ankylosis.[14,15].

Clinical evaluation and careful attention to radiological findings on regular follow-ups can help to pick up either disease at an early stage since it is not economical to perform HLA typing on every patient with arthritis.[13,16]

HLA-B27 is present in about 8-10% of the normal population and similarly can also be present in up to 10-11% of patients with RA, which thus probably represents a normal occurrence.[16] Thus, the HLA-B27 antigen can also be found in RA patients without any other features of AS. Rundback et al, attempted to identify distinguishing radiological features in the subset of RA patients who were HLA-B27 positive and found a 44% incidence of radiographic features of seronegative disease such as periostitis and sacroiliac joint abnormalities. Thus, they concluded that due to the wide range of radiological appearances, this subset of patients does not represent a distinct radiographic entity.[16] A patient with a negative RF and HLA-B27 may still have co-existence of both RA and AS. [13]

In almost all of the reported cases, AS preceded RA with a typical clinical scenario of a patient developing AS in his third or fourth decade followed by the occurrence of RA after about 15 years which develops initially as a peripheral polyarthritis.[7,17] This sequence is perhaps only natural since the mean age of presentation of AS is much earlier than that of RA. On the other hand, Toussirot and Acquaviva reported three patients with primary RA who subsequently developed AS The clinical presentation or the radiological features of either disease is not enhanced due to the presence of HLA-B27 in RA or HLA-DR4 in AS.[3]

Also, SI joint involvement in RA is less severe than that in AS. The coexistence of AS and RA could constitute an overlap syndrome akin to that which is seen in psoriatic arthritis.[3]

Conclusion

This combined entity requires accurate assessment or can be easily misleading. No definite conclusion is possible with regards to the etiopathogenesis of these conditions coexisting but this association with overlapping features and laboratory markers constitutes an interesting phenomenon and can pose a diagnostic dilemma.

Clinical Message

The occurrence of AS and RA is rare; and clinicians must remain suspicious of this entity while following up patients with any one diagnosis, as a diagnosis of one does not rule out the other. A patient with a negative RF and HLA-B27 may still have co-existence of both RA and AS. While laboratory test and clinical criteria are well-defined, simultaneous presence of concomitant symptoms can lead to a misdiagnosis and delay in initiation of appropriate pharmacotherapy. In addition, timely recognition and pharmacotherapy can possibly delay the aggravation of arthritis and delay the need for arthroplasty procedures in these patients.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given the consent for his/ her images and other clinical information to be reported in the journal. The patient understands that his/ her names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflict of interest: Nil **Source of support:** None

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