

A case of rheumatoid spondyloarthritis!

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Abstract

Coexistence of rheumatoid arthritis and spondyloarthritis in one patient is rare. Here we report a 42-year-old male with symmetrical small and large joint arthritis of upper and lower limbs, bilateral sacroilitis without clinical evidence of psoriasis or inflammatory bowel disease. He had high titres of both rheumatoid factor (RF) (125.5IU/ml) and anti-citrullinated peptide antibodies (ACPA) (>200U/mL) with human leukocyte antigen (HLA) typing revealing positivity for both HLA B27 and HLA DR4. Patient was managed with steroids, analgesics, Disease-modifying anti-rheumatic drugs (DMARDs) and biologicals. Our patient's initial clinical presentation and serology were suggestive of Rheumatoid arthritis but later he developed features of spondyloarthritis. Though he had genetic predisposition to both the diseases, clinical features and imaging studies on follow up were more in favour of spondyloarthritis. It is unclear as to whether the small joint arthritis of hands could be attributed to rheumatoid arthritis or spondyloarthritis. It is possible that these two conditions probably occur by chance or this could constitute an overlap syndrome between the two rheumatic conditions. Such cases pose difficulties in diagnosis and further study of such patients may help in early diagnosis and effective intervention.

Keywords: rheumatoid arthritis; spondyloarthritis; psoriatic arthritis

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Background

Rheumatoid arthritis and spondyloarthritis are two different musculoskeletal disorders with differences in etiopathogenesis, presentation and management. Occurrence of both these disorders in a single patient is rare. Further study of such patients would help in the prognostication and optimal management of these patients. Here we present a patient with clinical features suggestive of both Rheumatoid arthritis and Spondyloarthritis with genetic predisposition to both these conditions, posing a diagnostic challenge.

Case report

A 42-year-old male presented with complaints of pain and swelling involving predominantly small joints of upper limbs and large joints of lower limbs for 7 years with inflammatory back pain for

2 years. Pain and swelling initially involved small joints of upper and lower limbs with significant early morning stiffness and the symptoms were episodic being treated by a local practitioner with short courses of NSAIDs. Evaluation had revealed rheumatoid factor (RF) positivity, but patient was not on continuous treatment. Five years after the onset of his illness, the symptoms became persistent and he also developed inflammatory back pain. He had no history suggestive of ocular involvement, gastrointestinal disturbances, skin rashes or dandruff. He had no other co morbidities and was not a smoker or alcoholic. Family history was significant with a sibling suffering from rheumatoid arthritis.

Examination revealed a well built and adequately nourished male with normal cardiovascular, respiratory, neurological and abdominal examination findings. Skin, hair and nails were normal. Possibility of occult psoriasis was also ruled out after consultation with dermatologist. He had tenderness involving distal interphalangeal joints of both hands, right wrist swelling and tenderness of bilateral proximal interphalangeal joints and knee joints. Tests for involvement of sacroiliac joints like flexion abduction external rotation (FABER) test, pelvic compression and distraction tests were positive on both sides. Lumbar spine movements were not restricted.

Investigations revealed normal blood hemogram, urine routine, renal and liver function tests. Serum inflammatory markers like erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP) were both raised. Rheumatoid factor, anti-citrullinated peptide antibodies (ACPA) were both strongly positive. Human leukocyte antigen (HLA) typing revealed positivity for both HLA B27, HLA DR4. The results are summarised in Table 1.

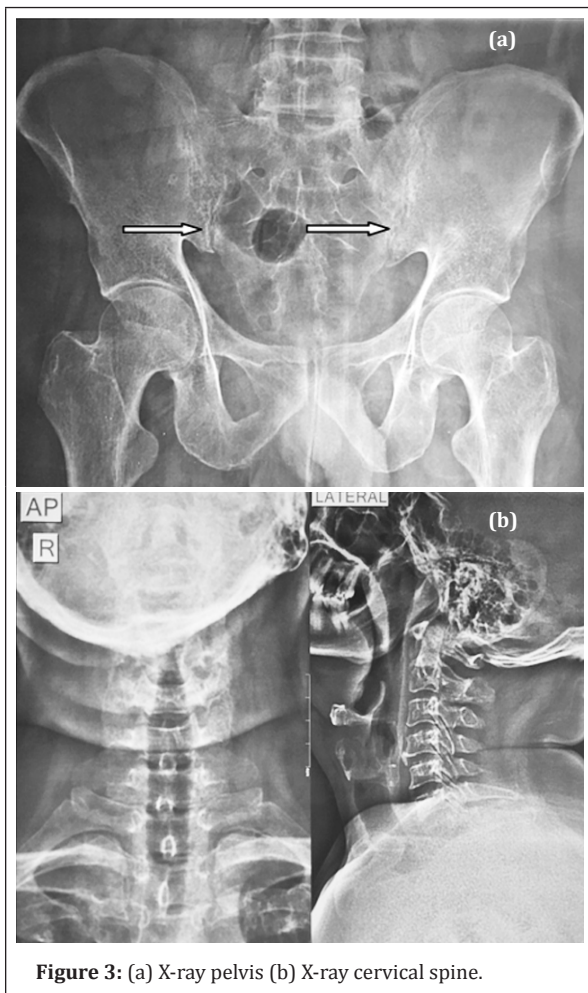
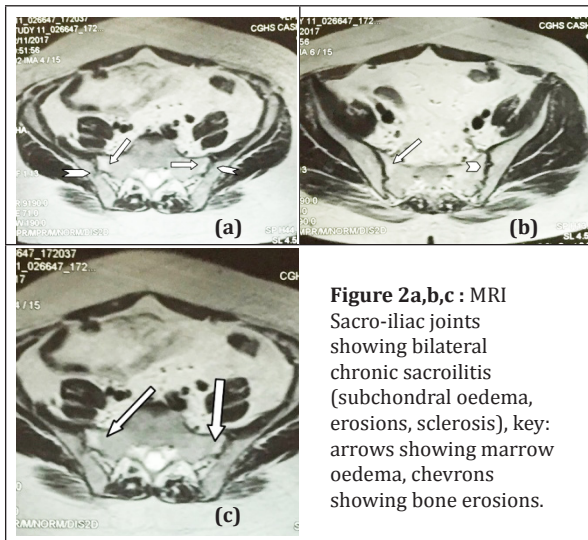
X-ray hands showed juxta articular osteopenia with periosteal reaction in the metacarpals and phalanges (Figure 1). Magnetic resonance imaging (MRI) showed bilateral chronic sacroilitis and mild osteoarthritic changes both hip joints with marginal osteophytes (Figure 2). X-ray of the pelvis showed erosions (depicted by arrows) in the sacroiliac joint and cervical spine revealed osteopenia with osteophyte at C4 vertebral level (Figure 3).

Table 1: Investigations.

Test name	Result
Hemoglobin	13 g/dl
Total white blood cell count	8600 cells/mm ³
Platelets	4.3lakh cells/mm ³
Blood Glucose	98 mg/dl
Serum creatinine	0.8 mg/dl
SGPT	23 IU/l
Serum Vitamin D	26.8 ng/ml
Serum Uric acid	6.1 mg/dl
Total cholesterol	145 mg/dl
Triglycerides	102 mg/dl
HDL	43 mg/dl
ESR	100 mm/hr
CRP	50.4 mg/L
HLA B27(by flowcytometry)	positive
HLA DR4(by Luminex)	positive
RF	125.5 IU/mL (Normal <30IU/mL)
ACPA	>200 U/mL (Normal <20 U/mL)



Figure 1: X-ray of hands – showing juxta-articular osteopenia and periosteal reaction.



Patient was treated with disease modifying anti rheumatic drugs (DMARDS) including hydroxychloroquine (HCQ), methotrexate and sulfasalazine nearly two years. In view of poor response to DMARDS, patient was started on injection

infliximab with good resolution of symptoms. The improvement in parameters is depicted in Table 2.

Table 2: Disease activity markers before and after 3 doses of injection infliximab.

Parameter	Before biological therapy	After biological therapy
ESR	100 mm/hr	44mm/hr
CRP	50.4 mg/L	4 mg/L
Tender joint count	26	6
Swollen joint count	8	0
BASDAI	6.6	2.4

Discussion

The coexistence of RA and ankylosing spondylitis (AS) is rare [1, 2] though there are several case reports of occurrence of both RA and AS in the same patient [1]. In the 1950's the term rheumatoid spondylitis was used to describe cases with features suggestive of both RA and ankylosing spondylitis [2]. According to Fallet et al. the probability of the two diseases in one patient is 1: 50 000 to 1: 200 000 [3].

Aside from the rarity of such a condition, it poses a diagnostic dilemma as the peripheral arthritis of ankylosing spondylitis is often indistinguishable radiologically, morphologically and histologically from RA [2]. Presence of rheumatoid nodules in such situations would be helpful as they are characteristic for RA, especially in sero-positive patients, whereas they are not observed in patients with AS [5].

RA is associated with HLA-DR1 and HLA-DR4 antigens and spondyloarthritis is associated with HLA-B27 but there is no association between RA and HLA-B27 [6]. Toussirot et al. noted that 6.6% of patients with RA may have positive HLA-B27, which is comparable to the general population (5-8%) and that the presence of HLA-B27 in RA does not influence the severity of RA and does not increase the incidence of sacroiliitis or enthesitis [4, 7, 8]. Reports of the influence of HLA DR4 on AS are however controversial with some reports suggesting that HLA DR4 does not have any influence on the course of AS [9, 10] and others noting that AS patients with HLA DR4 positivity have a 7-fold increased risk of peripheral arthritis [2].

Sacroilitis in RA is rare and spine involvement is usually confined to upper cervical spine. But, according to some reports, sacroilitis can occur in up to 20% of patients with RA [5]. Sacroilitis, when it occurs in RA usually involves the caudal aspect of the sacroiliac joint. And ankylosis, pseudo-narrowing of sacroiliac joints and reactive sclerosis are rare. Erosions are also ill defined [4].

RF occurs in 70–85% cases of RA [11]. But, its specificity is low. The frequency of RF increases with age and is present in conditions other than RA [12]. RF positivity in AS patients is rare and even if present, it is usually in low titres. In studies by Toussirot et al., RF was positive in 8.3% of patients with AS and in 9.8% of the control group, and this difference was not statistically significant and RF positivity in AS does not increase the risk of erosive form of peripheral arthritis [4]. Antibodies against citrullinated peptides are more specific for RA and are observed in 60–70% of patients with RA [12]. Though antibodies against citrullinated peptide antigens (ACPA) are specific for RA there are case reports of coexistence of RA and AS with positivity for both RF and ACPA [8].

Synovial biopsy and immunohistochemistry, although not done in our case, may be helpful in such cases for identification of the predominant disease phenotype.

In most patients with RA coexisting with AS described during the last decade, biological treatment with TNF inhibitor was administered [13, 14]. Our patient was managed on the same lines and though he was responding poorly to conventional synthetic DMARDs, he showed a good response to TNF inhibitor. Other treatment options include rituximab with Dundar et al. [15] describing a 63-year-old female patient with coexisting RA and AS treated with rituximab with good effect after failure of TNF treatment.

Conclusion

The typical patient with coexisting RA and spondyloarthritis, in studies, is a male developing axial spondyloarthritis at 20–30 years of age followed by peripheral arthritis a few years later [4] but this patient was unique in that, his presenting features were that of RA and that he had high titres of both RF, ACPA. It is probable that the coexistence of RA

and SpA in this patient occurred by chance. Though such cases pose a diagnostic dilemma, treatment can be tailored according to the predominant clinical manifestation with biological therapy being a good treatment option for cases, refractory to DMARDs and NSAIDs.

Conflict of interest

Authors declare no conflicts of interest.

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