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CASE REPORT

Remitting Seronegative Symmetrical Synovitis with Pitting Oedema (RS3PE) in a Young Patient Mimicking Syphilitic Arthritis: A Case Report.

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Abstract
Remitting seronegative symmetrical synovitis with pitting oedema (RS3PE) is a rare disorder characterized by acute symmetrical synovitis involving both hands and feet, accompanied by pitting oedema. The aetiopathogenesis of RS3PE is still obscured. It commonly occurs in individuals over 50 years of age and has a good prognosis. RS3PE responds well to corticosteroid. We report a case of a young male being treated for sexually transmitted disease (STD) who presented with recurrent episodes of bilateral leg oedema and symmetrical joint pain involving the ankles and hands. He was successfully treated with corticosteroid. Reactive arthritis was excluded in view of the recurrent episodes with no evidence of all phases of syphilis, especially tertiary syphilis.

Keywords: *Corticosteroid, Reactive arthritis, RS3PE, Sexual transmitted disease, Syphilis, Young.*

Introduction

RS3PE is a rare disorder of unknown aetiology and pathogenesis. It was first described in 1985 by McCarty [1]. Nevertheless, it has been suggested that autoimmunity with a possible association with human leukocyte antigen (HLA) haplotypes, plays an important role [2, 3]. RS3PE is characterized by pitting oedema and acute symmetrical synovitis of small joints of the hands, giving a boxing-gloves' appearance. RS3PE has been identified as a distinct entity that predominantly affects elderly male over 50 years of age [1, 4] and has been found to be associated with autoimmune diseases and paraneoplastic syndrome [5]. RS3PE is rare in young individuals, as previously reported [6]. We describe a a case of a young male who had been treated for sexually transmitted disease (STD) and presented with recurrent bilateral lower limbs pitting oedema and joint pain. He was tested negative for rheumatoid factor (RF), anti-citrullinated peptide antibody (ACPA), and other serological markers for autoimmune diseases, but responded well to corticosteroid.

Case report

A 34-year-old Indian man, who had been treated for STD (Venereal Disease Research Laboratory, VDRL 1:16, and positive Treponema Pallidum Hemagglutination Assay, TPHA) in a neighbouring country following unprotected sexual activity, presented with recurrent painless bilateral leg oedema for the past 4 years. He had no other medical illness. He smokes cigarettes and drinks alcohol occasionally but does not take any recreational drugs. He had mild abdominal distension without other gastrointestinal symptoms including melaena, hematemesis, and jaundice. He had no fever, rashes, neurological and cardiorespiratory symptoms. His urinary habit was normal with no cloudiness or frothiness of urine and there was no urethral discharge. He had no history of sore throat. Subsequently, he developed joint pain affecting the ankles and hands. There was no early morning stiffness.

Clinical examination revealed a well-built man, with tattoos on his arms. There were no signs of chronic liver stigmata, oral thrush, or abnormal dentition. Bilateral painless pitting pedal oedema, extending up to the knees and external genitalia, was present. The hands, ankle, and tarsal joints were swollen and tender. There was no deformity present. Peripheral lymph nodes were not palpable. Other systems were unremarkable.

Laboratory investigation revealed reactive VDRL with low titre 1:2, and positive TPHA. Biochemistry profile showed normal serum creatinine, serum albumin, absence of urinary protein, and normal 24-hour urine protein. The pro-B-type natriuretic peptide (BNP) level was normal. Serological markers rheumatoid factor (RF), anti-citrullinated peptide antibody (ACPA), anti-nuclear antibody (ANA), anti-double stranded DNA (anti-dsDNA), and complement levels (C3 and C4), were all negative. Anti-hepatitis C antibody, retroviral study and *Chlamydia* serology were also negative. Ultrasonography of the abdomen showed normal hepatobiliary and genitourinary systems. Computed tomography (CT) scan of the abdomen clarifies the unremarkable findings.

Based on the laboratory and imaging findings, a diagnosis of RS3PE was made and he was given prednisolone 20 mg daily, and spironolactone 25 mg daily. He was asymptomatic after a month of treatment without recurrence of oedema and joint pain. Both steroid and diuretic were then stopped and he remained in complete remission for more than 6 months. However, he is still indulging in sexual activity despite being advised against it.

Discussion

RS3PE is a rare distinct clinical entity of an autoinflammatory condition with an obscured aetiopathogenesis [1]. It has previously been associated with various rheumatic diseases, such as rheumatoid arthritis, systemic sclerosis, and systemic lupus erythematosus. The true

prevalence and incidence of RS3PE is unknown. However, it has been commonly reported among male Caucasians > 50 years of age which is one of the diagnostic criteria defined by Olive et al [7]. Other criteria include the presence of pitting oedema on the dorsum of both hands, sudden onset of polyarthritis, and negativity for RF. The peripheral oedema was not attributed to cardiac, liver, and renal aetiology in this patient.

This patient fulfilled the criteria except for age and achieved remission with a low dose of steroids. RS3PE is rare in young males. Nonetheless, three cases of RS3PE in young adults have been reported by Sattar [8].

Syphilis has been considered a great mimicker of other conditions due to its wide spectrum of clinical manifestations. Reactive arthritis is associated with gastrointestinal and urogenital infections, commonly due to *Chlamydia trachomatis*, *Salmonella*, *Shigella*, *Campylobacter*, and *Yersinia*. None of these were found in this patient. Syphilis, caused by the spirochete, *Treponema Pallidum*, does not typically cause reactive arthritis. Arthritis associated with tertiary syphilis was also absent in this patient. Nevertheless, syphilis had been reported to cause arthritis mimicking RA in a few cases [9] and resolved upon treating the syphilis. There were no cases of syphilitic arthritis associated with peripheral oedema or RS3PE specifically reported so far as illustrated by this patient. Neither any signs of all the three stages of syphilis showed in this patient.

Low dose steroids have been shown to be effective in achieving sustained remission in RS3PE patients, in addition to disease-modifying rheumatic drugs (DMARDs) e.g., hydroxychloroquine (HCQ)[1, 10].

Conclusion

RS3PE may occur in young age although more prevalent in elderly. Lack of definitive criteria for this rare and treatable condition may lead to underdiagnosis and delay in treatment. The diagnosis of RS3PE is by exclusion i.e., other differential diagnoses of rheumatological disorders should be thoroughly assessed.

Conflict of interest and financial disclosures
None.

Informed Consent
Written informed consent was obtained from the patient for the publication of this report and the accompanying images.

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Authors contribution
WS: Ideas, case management, data collection, and manuscript writing; DCL: case management and review of the manuscript.

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