CASE REPORT

REMITTING SERONEGATIVE SYMMETRICAL SYNOVITIS WITH PITTING OEDEMA

N.S. Neki, Ankur Jain, Rohit Bajaj, Mohit Manav Jindal

Department of Medicine, Government Medical College and Guru Nanak Dev Hospital, Amritsar, India

Remitting seronegative symmetrical synovitis with pitting oedema (RS₃PE) is a rare clinical entity often mimicking more common diseases like rheumatoid arthritis (RA) and polymyalgia rheumatica (PMR). Although the exact aetiology is not clear, yet it responds dramatically to low doses of steroids with excellent prognosis unlike RA and PMR. We report a case of 72 year old male agriculturist by profession presenting with acute polyarthritis and pitting oedema of both hands and feet as well as pretibial areas, diagnosed to be a case of RS₃ PE.

Keywords: Remitting, Seronegative, Synovitis, Elderly

J Ayub Med Coll Abbottabad 2014;26(4):628-9

INTRODUCTION

Remitting seronegative symmetrical synovitis with pitting oedema (RS₃PE) is a distinct clinical entity and HLA B7 positive. It mimics more common diseases like rheumatoid arthritis (RA) and polymyalgia rheumatic (PMR) and differentiation can be established by distinct clinical, radiological and immunological characteristics. The definite diagnostic criteria of RS₃PE include presence of seronegative acute polyarthritis, pitting oedema of hands and/or feet, old age, male dominance, excellent response to low dose steroids with long term remission. Hence it is a treatable condition, though rare. Although it is a benign condition, sometimes it may present as paraneoplastic manifestations of malignant disorders like non-Hodgkin lymphoma, chronic lymphatic leukaemia, pancreatic carcinoma, lung cancer, breast cancer, prostate cancer, bladder cancer, endometrial carcinoma and carcinoid.²⁻⁴ It can be associated with polyarteritis nodosa, pneumonia and amyloidosis.^{5,6} There is frequent association with knee synovitis and carpal tunnel syndrome.⁷

CASE REPORT

A 72 year old male farmer presented to the hospital with chief complaints of severe pain involving wrist, metacarpophalangeal and interphalangeal joints of both hands for 23 days. He was unable to make a complete fist. Local examination of joints revealed inflammation of flexor tendon sheaths of hands and feet along with tenderness on palpation. There were multiple tender swellings over the dorsum of hand and wrist as well as pitting oedema over dorsum of hands, feet and pretibial areas. The axial skeleton and proximal joints of the limbs were spared. General physical examination and systemic examinations were unremarkable. Laboratory investigations showed: haemoglobin 11.7g/dl, total leukocyte count 9200/cu mm, platelets 2.30 lac/cu mm, and ESR

85mm at the end of 1st hour. Differential leukocyte count (DLC) was N₆₅, L₃₂, M₂, E₁ Other laboratory investigations were: total serum proteins 6 gm/dl, serum albumin 3.8 g/dl, CRP 30 mg/ml, blood sugar fasting 90 mg/dl, blood urea 46 mg/dl, and serum creatinine 1.1 mg/dl. Rheumatoid factor and antinuclear antibody were negative. X-ray chest, thyroid profile, urine examination and 2D Echo were normal. X-ray of the joints showed no evidence of erosion. Ultrasonography of hands, wrist and feet revealed evidence of tenosynovitis involving flexor and extensor tendons. HLA typing of the patient was not done due to financial constraints. In view of the above findings, a diagnosis of remitting seronegative symmetrical synovitis with pitting oedema was made. The patient was put on low dose of steroids i.e., prednisolone 20 mg/day with enteric coated aspirin 352 mg given thrice daily after meals along with antacid, calcium and vitamin D supplementation. He showed dramatic improvement with subsidence of joints pain and oedema along with improvement of hand grip strength within 3 weeks of start of therapy. He was discharged after 4 weeks with no residual disability and normal ESR. Further follow up did not reveal any relapse.

DISCUSSION

MC Carty *et al* in the year 1985 first described RS₃ PE syndrome in 10 patients of which 8 were males and 2 females.⁶ He along with other colleagues Hunter & Russell in the year 1990 added 13 more cases to establish the clinico-radiological pattern of the disease.⁸ Now RS₃PE is discussed as a distinct clinical entity showing male predominance (M:F=2:1) with involvement of big and small joints of hands associated with tenosynovitis especially of flexor tendons of hands and accompanied by pitting oedema of dorsum of hands although oedema of feet or pretibial areas may or may not be observed.⁹ Kundu AK in his study¹⁰ has reported 5 cases occurring in old age showing significant improvement with the use of low dose steroids without

any regional disability. RS₃PE is characterized by acute onset of symmetric polysynovitis, pitting oedema of dorsum of hands and/or feet with negative rheumatoid factor and anti CCP along with positive inflammatory markers such as ESR and CRP. 6 Regarding aetiology of pitting oedema, it could be explained on the basis of marked extensor tenosynovitis as documented on MRI.¹¹ Agarwal et al¹² in their study have documented ultrasonography as a gold standard tool and most effective modality to diagnose tenosynovitis for evaluation of RS₃PE and they have also shown that extensor synovitis is much more common than flexor tenosynovitis as reported in our case. Tenosynovitis of both flexor and extensor tendons at the wrist and extensor tendons of the feet is the hall mark of RS₃PE as reported in the present case. Role of vascular endothelial growth factors has been postulated in the pathophysiology of RS₃PE.¹² Response to steroids is excellent as in our case. If there is poor or no response, one should think of underlying paraneoplastic syndromes^{1,2} since complete remission was observed in all cases after total resection of the tumour suggestive of para neoplastic syndrome. RS₃PE mimics RA and PMR but differs from them in the clinical, radiological and immunological characteristics. PMR is more frequent in females, involves shoulder and pelvic girdle with associated systemic symptoms, pitting oedema is rare, HLA-DR4, frequent relapses and recurrences; and responds only to steroids.

Hence differential diagnosis includes seronegative rheumatoid arthritis, acute sarcoidosis, ankylosing spondylitis, psoriatic arthropathy, mixed connective tissue disease, reflex sympathetic dystrophy, chondrocalcinosis, and amyloid arthropathy.

CONCLUSION

RS₃PE is a distinct rare clinical entity with excellent prognosis and dramatic response to low dose steroids unlike RA and PMR. It must be suspected in all

elderly patients presenting with acute polysynovitis and pitting oedema.

REFERENCES

- Dudler J, Gerster JC, So A. Polyarthritis and pitting oedema, Am Rheum Dis 1999;58(3):142–7.
- Sayarlioglu M. Remitting seronegative symmetrical synovitis with pitting oedema and malignancy. Eur J Gen Med 2004;1(2):3-5.
- Cobeta Garcia JC, D Omingo Morera JA, Martinez-Burqui J. Remitting seronegative symmetrical synovitis with pitting oedema (rs₃pe) syndrome, chronic lymphoid leukemia. Clin Exp Rheumatol 1999;17(2):266–7.
- 4. Goldenbing K, Rozenbaum M, Rosner I, Naschitz J, Kotler C. Remitting seronegative symmetrical synovitis with pitting oedema (RS₃PE) syndrome secondary to Non-Hodgkin lymphoma. Clin Exp Rheumatol 1998;16(6):767–8.
- Díez-Porres L, Muñoz-Fernández S, Aguado P, Alonso M, Martín-Mola E. Remitting seronegative symmetrical synovitis with pitting oedema as the first manifestations of psoriatic arthropathy. Rheumatology(Oxford) 2002;41(11):1333-5.
- Parisier KM, Canoso JJ. Remitting seronegative symmetrical synovitis with pitting oedema—Two cases of RS3PE syndrome. J Rheumatol 1991;18(8):1260–2.
- MC Carty DJ, O'Duffy JD, Pearson L, Hunter JB. Remitting seronegative symmetrical synovitis with pitting oedema (RS₃PE) syndrome. JAMA 1985;254(19):2763-7.
- Russel EB, Hunter JB, Pearson L, McCarty DJ. Remitting seronegative symmetric synovitis with pitting oedema 13 additional cases. J Rheumatol 1990;17(5):633–9.
- Kundu AK. Syndrome of remitting seronegative symmetric synovitis with pitting oedema (RS₃PE). J Assoc Physician India 2007;55:227–30.
- Cantini F, Salarani C, Oliveri I, Basozzil L, Macchioni L, Niccoli L. et al. Remitting Seronegative Symmetrical Synovitis with Pitting Oedema (RS₃PE) syndrome and a prospective follow up and magnetic resonance imaging study. Amer Rheum Dis, 1999, 58(4):230–6.
- 12. Aggarwal V, Dabra AK, Kaur A, Sachdev A, Singh R. remitting seronegative symmetric synovitis with pitting oedema (RS₃PE) syndrome. Ultrasonography as a diagnostic tool. Clin Rheumatol 2005;24(5):476–9.

Address for Correspondence:

Dr. N.S. Neki, Professor of Medicine, Govt. Medical College and Guru Nanak Dev Hospital, Amritsar, India, 143001.

Email: drneki123@gmail.com