

# REMITTING SERONEGATIVE SYMMETRICAL SYNOVITIS WITH PITTING OEDEMA SYNDROME, ASSOCIATED WITH PROSTATE ADENOCARCINOMA: A CASE REPORT

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## Abstract

Remitting seronegative symmetrical synovitis with pitting oedema (RS3PE) of the dorsum of the hands and/or feet can be observed in different inflammatory rheumatic diseases as well as in haematological and solid malignancies. McCarty *et al*, described this syndrome for the first time more than twenty years ago. Underlying malignancy should always be excluded in patients with RS3PE syndrome.

**Keywords:** RS3PE; Prostate Adenocarcinoma; Elderly.

## Introduction

Remitting seronegative symmetrical synovitis with pitting oedema (RS3PE) is a rare inflammatory type of arthritis that occurs predominantly in elderly men with clinical manifestations of acute onset pitting oedema of hands. Is usually characterized by a rapid-onset symmetrical synovitis; presence of pitting oedema of joints involved, especially the dorsum of hands; seronegativity of rheumatoid factor; absence of joint erosion; good response to low dose steroids with long term remission after withdrawal. In spite of appearing a well-characterized condition, subsequently research has shown that it is not a specific entity but rather a syndrome that can in fact represent the first manifestation of various types of rheumatic diseases and neoplastic conditions, usually of the elderly. In the last years, some reports described the RS3PE syndrome as paraneoplastic syndrome, exemplified in patients with distal extremity with swelling pitting oedema

as the first manifestation of haematological and solid malignancies. Herein we report a case of pitting oedema on hands in a patient with prostate adenocarcinoma.

## Case report

E.S., a 74-year-old male Caucasian patient, was admitted into the Rheumatology Clinic of our institution (Hospital Infante D. Pedro, E.P.E.) in the first week of January 2009 with pain and swelling of both hands with severe disability started suddenly during the two weeks before. The patient also had complaints of weight loss of 2kg in 15 days, epigastric pain and nocturia. His complaints did not decrease with nonsteroidal antiinflammatory drugs. No preceding history of major trauma, headache, fever, skin changes, anorexia, cough or intestinal disorders. His past medical history included two prostate surgical procedures, blood hypertension with no alcohol or tobacco abuse. The physical examination revealed swelling of both hands with pitting oedema noted over the dorsum (Figures 1 and 2) with inflammation of flexor tendon sheaths of the hands and swelling of knee joints. Laboratory data showed an erythrocyte sedimentation rate (ESR) of 130 mm/h and a C-reactive protein (CRP) level of 15.51 mg/dL. The haematological values and biochemical markers were found to be normal except for haemoglobin (10.6 g/dL) and albumin (2.6 g/dL). Rheumatoid factor (RF), anticitruline (anti-CCP), hepatitis B and C antibodies were negative. Serum prostate-specific antigen (PSA) level was raised (10.10 ng/dL with normal values under 4,0 ng/dL). Radiological examination of the hands showed soft tissue swelling and did not reveal any erosions. Chest radiograph and endoscopy of upper gastrointestinal system were normal. The diagnosis of remitting seronegative symmetrical synovitis with pitting oedema (RS3PE syndrome) was

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**Figure 1.** Pitting oedema of Dorsum of Hands before treatment



**Figure 2.** Oedema of Palm of Hands before treatment



**Figure 3.** Dorsum of Hands after Treatment



**Figure 4.** Palm of Hands after Treatment

suggested and, considering the possibility of an underlying malignancy, a prostate biopsy was performed. The patient was treated with oral diclofenac 150 mg/day and oral prednisolone 20 mg/day with an improvement of the hands oedema. Meanwhile prostate biopsy showed an adenocarcinoma and the patient underwent antiandrogenic therapy. There was remission of clinical findings (Figure 3 and 4) and improvement in laboratory parameters (Hb 12.4 mg/dL). The patient became independent in daily living activities.

## Discussion

In 1985, McCarthy *et al*<sup>1</sup> described a specific type of arthritis, with a sudden onset bilateral symmetrical synovitis of upper and/or lower limbs associated with a marked pitting oedema of the dorsum

of the hands (“boxing-glove” hand) and/or feet, especially affecting the elderly males and with an excellent prognosis. Etiology of the RS3PE syndrome is unknown but infectious agents or environmental factors are known to have impact in the development of this disease<sup>1</sup>. Serologic testes, like rheumatoid factor (RF) are negative and radiographic joint destruction is not seen<sup>1</sup>. The clinical findings originally described for RS3PE syndrome had also been observed in other diseases such as rheumatic diseases and neoplastic conditions of the elderly<sup>2</sup>. In fact, clinical findings of RS3PE syndrome were described in conditions like acute sarcoidosis<sup>3</sup>, and in other rheumatic diseases: polymyalgia rheumatica<sup>4,5</sup>, spondylarthropathies<sup>2,6</sup>, rheumatoid arthritis<sup>7</sup> and psoriatic arthritis<sup>8</sup>. Several articles have reported patients with distal extremity swelling with pitting oedema as the first manifestation of haematological and solid tumors<sup>9</sup>. Prostate, stomach and colon seem to be the most frequently in-

involved organs<sup>9</sup>, but association with non-Hodgkin lymphoma<sup>10</sup>, chronic lymphoid leukemia, T lymphoma<sup>11</sup>, hepatocellular carcinoma<sup>12</sup>, endometrial adenocarcinoma<sup>13</sup> or undifferentiated lung carcinoma<sup>14</sup> was also reported.

Patients with idiopathic RS3PE syndrome showed an excellent response to low doses of corticosteroids compared to the poor response of the RS3PE patients with associated neoplasia. However, in some reported cases RS3PE was relieved with steroid treatment before treatment of malignancy<sup>15</sup>. In our case, the patient received corticosteroids and hormone therapy almost in the same time. Therefore, the efficacy of steroids treatment should be questioned. A higher frequency of systemic signs and symptoms (fever, anorexia and weight loss) have been observed in patients with an underlying malignancy<sup>14</sup>.

In the clinical setting of RS3PE syndrome, the rheumatologist should consider malignancies in the absence of other associated rheumatic diseases, in the presence of systemic signs and symptoms and if the response to corticosteroids is poor<sup>5</sup>.

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