Inflammatory Polyarthritis in an 80-year-old Frail Male—A Case of Remitting Seronegative Symmetrical Synovitis with Pitting Edema (RS3PE)

Chang-Hung Chen¹, Horng-Ming Yeh²

Abstract

Sudden onset of inflammatory polyarthritis developed about six months ago in an 80-year-old healthy male. Sick and weak due to the inflammatory polyarthritis, the patient reported frailty symptoms including poor appetite, body weight loss, and inability to walk or standup without aids. On examination, pitting edema over both dorsal hands and feet was noted. Laboratory findings indicated normochromic normocytic anemia, raised inflammatory markers (ESR 44 /hr, CRP 10.22 mg/dl), and normal WBC while tests for autoantibodies, rheumatoid factor and anti-cyclic citrullinated protein antibody were all negative. Radiological findings of the extremities showed swollen soft tissue without any erosion. Detection of tumor markers was unremarkable, and monoclonal spike was not observed in serum protein immunoelectrophoresis. Remitting seronegative symmetrical synovitis with pitting edema syndrome (RS3PE) was diagnosed based on the findings of bilateral pitting edema of hands, sudden onset of polyarthritis, an age over 50 years old, and seronegative rheumatoid factor in this patient after excluding other possibilities. This patient showed an excellent response to low doses of corticosteroids. RS3PE should be considered for elderly male patients with acute onset of polyarthritis and pitting edema of extremities.
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Key words: remitting seronegative symmetrical synovitis with pitting edema syndrome, inflammatory polyarthritis, frailty

¹Department of Internal Medicine, ²Department of Geriatrics, Tainan Municipal Hospital
Correspondence to: Horng-Ming Yeh
No.670, Chung-Der Road, Tainan 701, Taiwan
Tel: (886) 6-2609926
E-mail: yehwu@ms31.hinet.net
Introduction

Remitting Seronegative Symmetrical Synovitis with Pitting edema (RS3PE) syndrome is a subset of acute onset polyarthritis which mainly affects elderly men with clinical manifestations of pitting edema of the hands [1]. If the diagnosis of polyarthritis is delayed, inflammation will make elderly patients weak and sick. In this case report we discussed a previously healthy 80-year-old male who had inflammatory polyarthritis. Poor appetite, body weight loss, inability to walk or standup without aids comprised the frailty symptoms, which were significantly alleviated after appropriate diagnosis and treatment.

Case presentation

An 80-year-old male patient was admitted in February 2011 with a 6-month history of painful swollen hands and knees associated with worsening mobility. The symptoms were acute in onset, non-traumatic and marked with an absence of any associated features of connective tissue disease. He was healthy without systemic disease and could do exercise such as push-ups and sit-ups every day before this episode. Sudden onset of bilateral proximal interphalangeal (PIP) joints, metacarpal phalangeal (MCP) joints, shoulder, knee, ankle pain and swelling developed about six months ago. He had visited neurologic, orthopedic and Chinese medicine departments for help but in vain. Inflammatory polyarthritis made him sick. The frailty symptoms in this patient included poor appetite, body weight loss, inability to walk or standup without aids. Arthralgia restricted his mobility while poor appetite caused inadequate nutrition, making him weak and eventually resulting in loss of body weight.

On examination, pitting edema over both dorsal hands and feet was noted. He also had synovitis at PIP, MCP joints, wrists and effusion of both knees and ankles. Laboratory findings included hemoglobin of 10 gm/dl with normochromic normocytic anemia, raised inflammatory markers (ESR 44 /hr, CRP 10.22 mg/dl), and normal WBC. Tests for autoantibodies, rheumatoid factor and anti-cyclic citrullinated protein antibody (anti-CCP) were all negative. Radiological findings of hands, ankles and knees show swollen soft tissue with no erosion. Chest X ray was negative, and abdomen sonogram showed parenchymal liver disease only. The patient was also screened for associated malignancies, and tumor markers including CEA, AFP, CA19-9 and PSA appeared to be normal. Serum protein immunoelectrophoresis showed no monoclonal spike. Gallium whole body tumor scan revealed...
inflammation over upper jaw only.

A diagnosis of remitting symmetrical seronegative synovitis with pedal edema was suggested, and patient responded extremely well to solucortef 100mg st and low dose of prednisolone at a daily dose of 7.5 mg. The inflammatory markers decreased (ESR 38/hr, CRP 1.81 mg/dl) after ten weeks of treatment. Five months of follow-up with tapering of prednisolone showed complete recovery without any flare-ups.

**Discussion**

RS3PE syndrome is a rare inflammatory disease, first described by McCarty et al. in 1985 [1]. This syndrome is usually found in elderly patients and characterized by acute onset, symmetrical synovitis, pitting edema over dorsal hands and feet, seronegativity for rheumatoid factor, and excellent response to low-dose corticosteroid therapy [2]. Sudden onset of bilateral PIP, MCP, shoulder, knee, ankle joints pain and swelling developed about six months ago in this previously healthy 80-year-old man. Excellent response was observed after the initiation of low dose corticosteroid therapy, a result consistent with the above criteria.

Although the pathogenesis of RS3PE remains unknown, over-production of inflammatory cytokine IL-6 along with vascular endothelial growth factor (VEGF), matrix metalloproteinase-3, and serum amyloid A has been demonstrated to contribute to its development [3-5]. The diagnostic delay in this patient caused prolonged inflammatory status, and several syndromes of frailty, notably poor appetite, body weight loss, and weakness in getting up, were noted in this patient.

Frailty as physiologic vulnerability and progressive decline is likely to be multifactorial in etiology. The physiologic alterations that have been associated with frailty are complex, and inflammation may be one of the factors increasing the risk of frailty [6]. Fried et al characterized frailty as the presence of three of five central components: unintentional weight loss, slow walking speed, self-reported exhaustion, low energy expenditure, and grip weakness [7].

There have been 32 case reports of cancer in RS3PE since 1985 in a total of 59 patients with RS3PE [8]. In contrast to RS3PE without malignancy, those with malignancy tend to respond poorly to glucocorticoid therapy and to have more dramatic systemic symptoms [9]. The dramatic response to low-dose corticosteroid therapy and negative tumor screening study precluded the possibility of malignancy in this patient.

Differential diagnosis led to polymyalgia rheumatica, rheumatoid arthritis, late onset
spondyloarthropathy, mixed connective tissue disease, chondrocalcinosis and amyloid arthropathy [10]. In this male patient, shorter natural course, increased acute phase reactants, lack of bony erosion, and remarkable sensitivity to low dose prednisolone supported the diagnosis of RS3PE syndrome.

As the population aged over 65 years continues to grow, recognition of frail elderly adults becomes increasingly important in the implementation of targeted intervention and medical care.

RS3PE should be considered for elderly frail patients with acute onset of polyarthritis and pitting edema of the extremities without an underlying systemic cause.

References


一位80歳衰弱老人的發炎性多發關節炎—
緩解性血清陰性對稱型滑膜炎併壓陷性
水腫症候群個案報告

陳長宏¹ 葉宏明²

摘　要

一位80歳老人大約六個月前突然發生多處發炎性關節炎，使得原本健
康良好的他變得病弱。衰弱的症狀包含胃口不佳、體重減輕，無法獨立起
立及行走。檢查時發現，病人兩側手背踝背有壓陷性水腫，實驗室檢查發
現正球性貧血，發炎指數升高 (ESR 44/hr, CRP 10.22 mg/dl)，白血球數正
常。自體抗體檢查、類風濕因子、抗環瓜氨酸抗體皆為陰性。雙手、踝關
節及膝關節X光檢查發現軟組織水腫但無侵蝕現象。腫瘤標記正常，血中蛋
白質免疫電泳檢查也正常。根據此病患的雙手壓陷性水腫、突發性多處關
節炎，年紀大於50歲，類風濕因子陰性，並排除其他可能疾病後，診斷為
緩解性血清陰性對稱型滑膜炎併壓陷性水腫症候群，此病患接受低劑量類
固醇之後反應極佳。當老年病患罹患突發性多處關節炎及壓陷性水腫時應
考慮此病。
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關鍵詞：緩解性血清陰性對稱型滑膜炎併壓陷性水腫症候群、發炎性多發
關節炎、衰弱