Triggering Role of Trauma on Initiation of SAPHO Syndrome: A Case Report.

SAPHO Sendromunda Travmanın Tetikleyici Rolü: Bir SAPHO Olgusu Nedeniyle

Ümit Dinçer[1], Barış Nacır[2], Engin Çakar[1], Mehmet Zeki Kıralp[1], Hasan Dursun[1]
[1]Gülhane Askeri Tıp Akademisi Haydarpaşa Eğitim Hastanesi, Fiziksel Tıp ve Rehabilitasyon Kliniği, İstanbul, Türkiye
[2]Gümüşsuyu Asker Hastanesi, Fiziksel Tıp ve Rehabilitasyon Kliniği, İstanbul, Türkiye

Abstract
SAPHO is a clinical syndrome characterized by Synovitis, Acne, Pustulosis, Hyperostosis, and Osteitis. Since the time it was first described, it has been diagnosed at an ever increasing rate. Although cases associated with palmoplantar pustulosis (PPP) have been reported, the relationship of the SAPHO syndrome and spondyloarthropathies with trauma remains poorly understood. While there are case reports in the literature which describe spondyloarthropathies following physical trauma, information exists indicating that psychological trauma and stress can also trigger or exacerbate palmoplantar pustulosis and psoriasis. Our case had PPP which arose 3 months after experiencing trauma on her shoulder and neck regions. In addition, she had low back pain and anterior chest wall pain which appeared after a short time following the onset of PPP. Unilateral sacroiliitis and bilateral sternoclavicular joint involvement were established in the patient. *(Romatizma 2008; 23: 106-8)*

Key words: SAPHO, palmoplantar pustulosis, trauma, psoriasis

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Özet
SAPHO sendromu Sinovitis, Akne, Pustulosis, Hiperostozis ve Osteitis ile karakterize bir sendromdur. Tanımlandığı zamandan beri artan bir oranda tanı konulmaktadır. Spondiloartropatiler ve SAPHO sendromu ile palmoplantar püstülozis (PPP) sendromu arasında bir ilişkinin varlığı bilinmekle birlikte, bu hastalıkların travma ile ilişkisi henüz tam olarak anlaşılamamıştır. Literatürde fiziksel travmayı takiben gelişen spondiloartropati olgularının yanısıra, fiziksel ve psikolojik travmalar ile tetiklenen veya alevlenen PPP ve psöriazis olguları mevcuttur. Bizim olgumuzda hırsızlık maksadıyla uğradığı bir fiziksel saldırı ile omuz, boyun bölgelerinden travmatize olmuş, 3 ay sonra başlayan PPP ortaya çıkmış ve kısa süre sonra bel ve ön göğüs duvarı ağrısı ilave olmuştur. Klinik araştırma sonrasında unilateral sakroiliit ve bilateral sternoklavikular eklem tutulumu olduğu belirlenmiştir. *(Rheumatism 2008 23: 106-8)*

Anahtar Sözcükler: SAPHO, palmoplantar pustulozis, travma, psoriasis

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Introduction

SAPHO syndrome is a member of spondloarthropathies (SpA) which is characterized by Synovitis, Acne, Pustulosis, Hyperostosis, and Osteitis (osteomyelitis). In 1987, Chamot et al. published a typically descriptive case characterized by synovitis, acne, pustulosis, hyperosteosis, and osteitis (1). PPP is characterized by sterile pustules, erythema and scaing. The lesions often begin unilaterally on the palm or sole and later spread to both palms and soles. The distribution of the lesions is then mainly symmetrical (2).

Case

A 38-year-old female patient. She was a secretary and single. She had not any family and personal history of arthritis, skin disorder, genitourinary and gastrointestinal tract infections. She had developed shoulder and back pain after experiencing a physical traumatic event (insult as an attempt of robbery). Pustular lesions in her hands and feet had occurred 3 months later. The condition had been dianosed as pustular psoriasis. Pain on the anterior chest wall as well as swelling of right sternoclavicular joint had deveoped along with the deterioration of the current

Address for Correspondence/Yazışma Adresi: Dr. Ümit Dinçer, Gülhane Askeri Tıp Akademisi Haydarpaşa Eğitim Hastanesi, Fiziksel Tıp ve Rehabilitasyon Kliniği, İstanbul, Türkiye
Tel.: +90 216 418 40 03 E-posta: drumitdincer@yahoo.com

lesions. Stiffness lasting for 2 hours in the morning with back pain had been present during the last 3 months. Mobilization of the lumbar vertebrae was painful and 1/2 restricted. Sacroiliac stretch tests were positive particularly on the right side. Schober test was 3 cm. The right sternoclavicular joint was swollen, and extremely tender on pressure, the left sternoclavicular joint was painful on pressure, but not swollen. There were diffuse pustular lesions on palmar sides of both hands, and also on plantar sides of the feet. Pustular lesions were noninfectious, and they were appearing and disappearing within two weeks. No psoriatic or pustular lesions were present on the scalp, or on the other regions of the skin. Cutaneous lesions were severely pruritic occasionally. (Figure 1.) Erythrocyte sedimentation rate (ESR) was 53 mm/h (range 0-20 mm/h) , CRP level was 65.8 mg/dL (normal value < 5 mg/dl, and RF was 7,1 IU/ml (normal value < 2 IU/ml). Serum Calcium, CPK, ALP, Parathyroid Hormone levels were normal. HLA B27 was negative. Possible focus of infections were investigated but were not isolated any fungal and bacterial agents. In radiographic evaluation, scleroses as well as narrowing of iliac wing of the right sacroiliac joint was present. (Figure 2). Erosions and cortical irregularity were detected in plain graphies of both sternoclavicular joints (SCJ). In tomographic examination of the sternoclavicular joint, cortical irregularity in sternum and clavicle were present. Involvements showing activity suggestive of arthritis in bilateral sternoclavicular joints were present in whole body bone scintigraphy with 20 mCI Tc99m- MDP (Figure 3).

Discussion

The relationship between trauma and spondyloarthropathy is still unclear although many case reports and a few clinical trial have suggested trauma might initiate or trigger the development of SpA. We aimed in this report to speculate the relationship between trauma and SAPHO. The factor or factors which trigger the onset of the SAPHO is not precisely identified yet. Even so, as in our case, there are views as well as findings which suggest that physical and psychic traumas trigger the development of spondyloarthropathies such as Psoriatic arthritis (2-6), Reiter's Syndrome (7,8), and ankylosing spondylitis (5,7,9). Nonetheless, no well-documented information is available on whether exclusively the joint which is exposed to trauma is involved alone or not. While the etiology of SpA's (included SAPHO) has not been wholly identified yet, whether trauma represents a widespread and precise etiologic factor is still unclear (2-4). Yet, in this case, the presence of complaints beginning after exposure to psychic and physical trauma brings about some doubts on whether this situation might be a casual event. The patient has initially developed lesions in her palms and feet later accompanied by back pain and pain in the anterior chest wall notwithstanding the improvement of her shoulder and neck pain within a month following the trauma. In this case, we believe that physical trauma might have played a role as a triggering factor. Likewise, in a family study which demonstrated the characteristics of SAPHO syndrome, Dumolard et al. presented three cases each of whom had a history of a traumatic event before onset. They denote that the physical traumas being experienced shortly before the onset of the illness represent the prominent features of the condition, and submit their strong belief that the traumatic event might have played a key role on the beginning process of the illness (10). Further, Sandorfi et al. presented four cases in which they suggested an association with trauma. In three of these cases, psoriatic arthritis was present which was triggered and precipitated by trauma, and spondyloarthropathy developing immediately after a traumatic event was present in one case (5). In a retrospective study, 288 patients with spondyloarthropathies were assessed, and 12 (4.2%) of them were found as having a history of trauma, and they underlined that this condition might be an important factor which triggered SpA (11). Concordant with the literature, the arthritis which aroused in this case have developed following a trauma, and the subsequent etiologic research have revealed no pathogenic agent, or any other origin. However, by which means trauma triggers the immunologic mechanisms occurring in the synovial tissue and the remaining articular components is not wholly understood. Trauma was defined by Jun et al. as physical trauma occurring within 1 month before the articular symptoms began (11). In our case, the gap between the trauma and PPP was 3 months and she had back pain and morning stiffness in this time. However the patient was not investigated for evaluation back pain and spondyloarthropathy. In study comprising 300 patients with Psoriatic arthritis, Punzi et al. (12) reported that 8% of

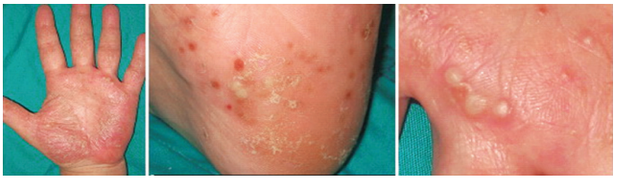

Figure 1. Cutaneous lesions on palm and sole

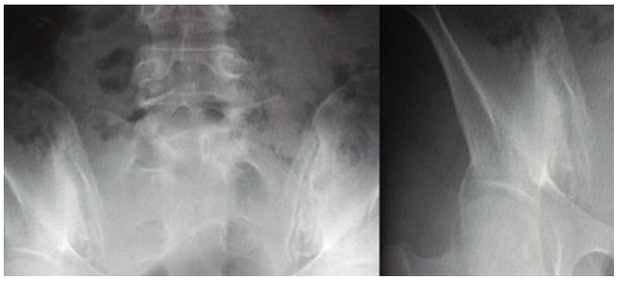

Figure 2. Radiograph of the sacroiliac joint shows sclerosis and narrowing of iliac wing of the right sacroiliac joint.

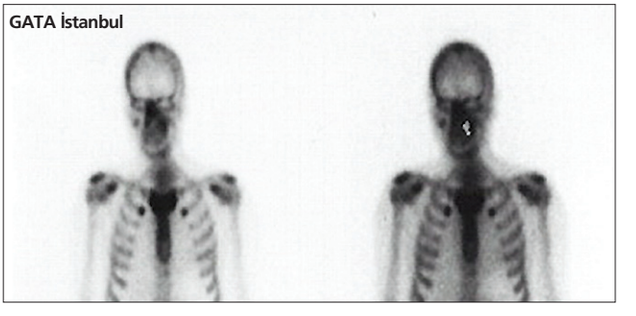

Figure 3. Bone scintigraphy with 20 mCI Tc99m- MDP shows activity uptake of bilateral sternoclavicular joint.

the patients had a history of trauma 3 months or shorter than 3 months prior to the onset of the disease (post-traumatic Psoriatic Arthritis). Palmoplantar pustulosis is a variant of psoriasis, and the role of trauma and emotional stresses which are known as the factors triggering and precipitating psoriasis is generally accepted.

SAPHO syndrome is clinically heterogeneous, covering several disease. Definite and correct diagnosis is hard and some clinical, radiological and histopathological signs are required. Since 1986, two new diseases belonging to SAPHO syndrome as nosological entities were documented: Pustulo-psoriatic hyperostotic spondyloarthritis (PPHS) and chronic recurrent multifocal osteomyelitis (CRMO). SAPHO syndrome and their variants should be recognized from ankylosing spondylitis, osteomyelitis, chronic polyarthritis, benign or malign bone tumors and psoriatic spondylarthritis (13). Diagnosis of SAPHO is so hard because of some pitfalls. Most important pitfalls are misdiagnosis and inattentiveness. Despite the limited knowledge from the literature, findings exist that suggest a relationship between cases with PPP as well as spondyloarthropathy-Psoriatic arthritis and emotional and/or physical trauma. However, it is clear that additional cases and further investigations are required for achieving stronger evidence.

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KONGRE TAKVİMİ

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