



## Complex Regional Pain Syndrome After Herpes Zoster: A Case Report

### Herpes Zoster Sonrası Kompleks Bölgesel Ağrı Sendromu: Olgu Sunumu

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### Abstract

A 78-year-old male patient, diagnosed with herpes zoster infection, had color change, stiffness, swelling and burning pain on his left hand. Hand joints were painful, joint range of motion decreased and hyperpigmented, macular lesions on the left C5-C6 dermatome region was determined. After the medical and physical therapy programme, symptoms decreased significantly. Complex regional pain syndrome (CRPS) is a painful disorder with swelling, stiffness in joints, vascular instability, and dystrophic skin changes. Symptoms most commonly occur after trauma, stroke, surgery, myocardial infarction, fracture, cancer. In this case, an unusual cause of complex regional pain syndrome, herpes zoster, was reported. Only a few cases have been reported to date. In this case report, it is emphasized that CRPS can occur as a complication of many conditions. Early diagnosis and appropriate treatment lead to better outcomes.

**Keywords:** Complex regional pain syndrome, herpes zoster, rehabilitation

### Öz

Herpes zoster enfeksiyonu tanılı 78 yaşında erkek hastanın sol elinde renk değişikliği, tutukluk, şişlik ve yanıcı ağrısı mevcuttu. El eklemleri ağrılı, eklem hareket açıklığı azalmış ve hiperpigmente, sol C5-C6 dermatomal bölgede maküler lezyonlar tespit edildi. Medikal ve fizik tedavi programı sonrası semptomlar anlamlı şekilde azaldı. Kompleks bölgesel ağrı sendromu (KBAS) şişlik, eklemlerde tutukluk, vasküler instabilite, distrofik deri değişiklikleriyle ağrılı bir bozukluktur. Semptomlar sıklıkla travma, inme, cerrahi, miyokard enfarktüs, kırık, kanser sonrası görülür. Bu olguda kompleks bölgesel ağrı sendromunun olağandışı bir nedeni olan herpes zoster bildirilmiştir. Bugüne kadar sadece birkaç olgu rapor edilmiştir. Bu olgu sunumunda KBAS'nin birçok durumun komplikasyonu olarak görülebileceği vurgulanmaktadır. Erken teşhis ve tedavi, olumlu sonuçlara yol açmaktadır.

**Anahtar kelimeler:** Kompleks bölgesel ağrı sendromu, herpes zoster, rehabilitasyon

### Introduction

Complex regional pain syndrome (CRPS) (reflex sympathetic dystrophy) is a painful disorder that affecting the hands but also arms, legs and limbs. The clinical features are spontaneous pain, hyperalgesia, stiffness, impairment of motor function, swelling and autonomic abnormalities. Symptoms most commonly occur after trauma. Other causes include infection, stroke, surgery, myocardial infarction, fracture, cancer (1). Although herpes zoster was first described by Sudeck (2) as a complication in 1901, only a few reports of herpes zoster as the cause of this syndrome has been reported (3-7). We describe this case of a patient with CRPS features after an herpes zoster infection.

### Case Report

A 78 years old male patient, with a vesicular rash that is limited to C4-C5-C6 dermatomes, was diagnosed with herpes zoster

infection in dermatology clinic. The patient had been treated 3000 mg/day valacyclovir for ten days. After ten days, swelling of the dorsum of the left hand and burning pain in the hand and fingers evolved gradually. The patient had gabapentin, tramadol and nonsteroidal anti-inflammatory drugs (NSAID) therapy, but symptoms increased. The patient was seen in our clinic three months later, with burning pain in the left hand and fingers, stiffness in the fingers, decrease of nail growth. He had no systemic disease. In physical examination, hyperpigmented, macular lesions in on the left C5-6 dermatome, blue colored cold skin and dryness in on the hand, fragile nails were seen (Figure 1). The wrist, metacarpophalangeal, distal and proximal interphalangeal joints were painful and decreased range of motion were determined. All of the laboratory test values (hemogram, biochemistry, sedimentation, C-reactive protein, rheumatoid factor, thyrotrophin-stimulating hormone levels) were in normal limits, only 25 hidroxy vitamin D3 level was

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22 ng/mL. Patchy osteoporosis were seen in the radiographs on the left hand (Figure 2). The patient was diagnosed as CPRS after herpes zoster and given Vitamin D3 1000 IU/per day, pentoxifylline 400 mg/day, pregabalin 75 mg/day, acetaminophen 90 mg/day. Physical therapy was planned for 30 sessions, which included contrast bath, left hand, wrist transcutaneous electrical nerve stimulation, stretching and strengthening exercise. Six weeks later, the complaints of the patients were markedly resolved and, dryness, blue color of skin were disappeared (Figure 3).

Written informed consent was obtained from the patient.

## Discussion

Herpes zoster is a self-limiting disease, with pain quenching at the end of vesicular eruption in dermatomal distribution.



**Figure 1.** Hyperpigmented, macular lesions in on the left C5-6 dermatome, blue colored cold skin and dryness in on the hand, fragile nails are seen



**Figure 2.** Patchy osteoporosis are seen in the radiographs on the left hand

Herpes zoster results from reactivation of latent varicella-zoster virus within the sensory ganglia (8-10). The incidence and severity of herpes zoster increase with advancing age and immunodeficiency or cancer (11); more than half of all persons are older than 60 years. Also our patient was 78 years old, had no systemic disease. The most frequent debilitating complication is postherpetic neuralgia, a neuropathic pain syndrome that persists or develops after the dermatomal rash has healed (9,10,12). Other neurologic complications are peripheral motor neuropathy, cranial nerve palsy, myelitis, encephalitis, cerebral thrombotic vasculopathy, acute polyradiculitis and aseptic meningitis (13,14). CRPS is a rare complication and only a few reports have been described in the literature (3-7). A case was a 65 years woman with characteristic signs and symptoms of CRPS in the right upper limb. CRPS was appeared four weeks after a herpes zoster infection. Intranasal calcitonin and physiotherapy lead to progressive functional and pain improvements (3). Similarly to the previous case, a 64-year-old woman with CRPS in the right hand that appeared four weeks after she had a herpes zoster infection, had medical treatment (diclofenac sodium, diltiazem, gabapentin, and lansoprazole) and physical therapy. She achieved a progressive improvement with early diagnosis and treatment (4). In our patient, CRPS development time was shorter from above cases. Despite different treatment options, progressive improvements were obtained in all studies. Several hypothesis can explain the mechanism of herpes zoster in causing CRPS, the first mechanism is, herpes zoster causes intense pain. This initial afferent nociceptive stimulus can sensitize multiple sympathetic neurons, resulting sympathetic outflow. The second mechanism is secondary inflammation due to cytopathic changes of herpes zoster infection. The third mechanism is spontaneously abnormal synapses between the efferent sympathetic nerves and afferent sensory nerves due to herpes zoster infection (15-17). Specific criteria for the diagnosis of CRPS were adopted in 2013 as the new international standard by the International Association for the Study of Pain (Table 1) (18). A comprehensive, integrated multidisciplinary



**Figure 3.** The hands of the patient six week after the treatment

**Table 1. Research diagnostic criteria for complex regional pain syndrome**

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|---|
| Continuing pain, which is disproportionate to any inciting event  |
| At least one symptom in three of the four following categories*:<br>Sensory: Hyperalgesia and/or allodynia<br>Vasomotor: Temperature asymmetry and/or skin color changes and/or skin color asymmetry<br>Sudomotor/edema: Edema and/or sweating changes and/or sweating asymmetry<br>Motor/trophic: Decreased range of motion and/or motor dysfunction (weakness, tremor, dystonia) and/or trophic changes (hair, nails, skin)   |
| At least one sign at time of evaluation in two or more of the following categories*:<br>Sensory: Evidence of hyperalgesia (to pinprick) and/or allodynia (to light touch or deep somatic pressure, or joint movement)<br>Vasomotor: Evidence of temperature asymmetry and/or skin color changes and/or asymmetry<br>Sudomotor/edema: Evidence of edema and/or sweating changes and/or sweating asymmetry<br>Motor/trophic: Evidence of decreased range of motion and/or motor dysfunction (weakness, tremor, dystonia) and/or trophic changes (hair, nails, skin) |
| There is no other diagnosis that better explains the signs and symptoms   |

treatment that includes medical, psychological, and physical and occupational therapy is needed in the treatment of CPRS. Randomized trials suggest that steroids, NSAID, opioids, immun modulators, analgesic antidepressants, bisphosphonates, calcitonin, anticonvulsants, NMDA receptor antagonists, calcium channel blockers, block therapies, surgical sympathectomy, and spinal cord stimulation may be effective treatments (18,19). In our patient, oral and topical NSAIDs, anticonvulsants, physical therapy and Pentoxifylline was used. Physical therapy increases patients range of motion, flexibility and strength (20). NSAIDs are used to treat pain plus inflammatory involvement in CRPS (21). Most often used as anticonvulsants, several have efficacy in neuropathic pain (22,23). Also Pentoxifylline, a cytokine inhibitor, was used in our treatment to reverse nociceptive sensitization and vascular abnormalities (24). It is clinically accepted that early diagnose and treatment in CRPS will lead to better outcomes. Also our patient, showed a progressive improvement with early medical treatment and physical therapy. Although CPRS is a self limited clinical course in most cases, some patients may progress for years leading a major functional disability of the affected extremity. The early management of this clinical entity is very important in daily clinical practice.

### Ethics

**Informed Consent:** Written informed consent was obtained from the patient.

**Peer-review:** Externally and internally peer-reviewed.

### Authorship Contributions

Surgical and Medical Practices: A.E., F.B., Concept: A.E., F.B., S.Ö., Design: A.E., F.B., S.Ö., Data Collection or Processing: F.B., Analysis or Interpretation: F.B., S.Ö., Literature Search: F.B., Writing: F.B., S.Ö.

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