

# COMPLEX REGIONAL PAIN SYNDROME-LIKE SYMPTOMS FOLLOWING HERPES ZOSTER INFECTION WITH DISTAL EXTREMITY INVOLVEMENT: A CASE REPORT

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**Background:** Complex regional pain syndrome (CRPS)-like symptoms are rarely observed after herpes zoster (HZ) infection. Especially in the presence of a rash spreading to the distal extremity, CRPS-like symptoms are more common.

**Case Report:** In this report, we present a 67-year-old patient who developed CRPS-like symptoms along with persistent severe pain after HZ infection, whose symptoms resolved after sympathetic ganglion blockade.

**Conclusions:** CRPS-like symptoms are rarely to occur after HZ infection, especially in patients with HZ infection affecting the distal extremity. In these cases, early interventional procedures may be effective in resolving clinical symptoms, may affect prognosis, and may help prevent persistent pain.

**Key words:** Complex regional pain syndrome, herpes zoster, varicella zoster, postherpetic neuralgia

## BACKGROUND

Herpes zoster (HZ) is usually a self-limiting infection with unilateral vesicular eruptions, but neurological complications can be seen, especially in the elderly (1). Although neurological complications, such as postherpetic neuralgia (PHN), encephalitis, meningitis, myelitis, and monoparesis, following HZ infection are well known (2), complex regional pain syndrome (CRPS)-like symptoms have been rarely reported (3-7). The mechanism and frequency of CRPS-like symptoms after HZ are unknown, and a limited number of cases reported in the literature have been reported in patients with distal extremity rash. In this case report, we demonstrated the successful treatment of a patient with distal extremity HZ rash who developed CRPS-like symptoms after infection with sympathetic block. We aimed to emphasize

the prognosis effect of early intervention in these cases, which are very rarely reported causes of resistant pain in the literature.

## CASE REPORT

A 67-year-old male patient presented with severe pain in the right shoulder and forearm.

The patient, with a past medical history significant for diabetes mellitus, presented to our clinic with reports of ongoing severe pain following HZ infection 3 months prior. On physical examination, there were diffuse hyperpigmented macular scarring areas on the right forearm and hand spreading to the right C5-C6 dermatome, along with skin discoloration on the right hand, dystrophic nail changes, edema and temperature increase in the right hand, allodynia, and hyperalgesia.

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The patient received oral acyclovir and prednisolone treatment during the acute herpes infection period, and was receiving tramadol 400 mg/g and pregabalin 450 mg/g treatment for pain. Given the patient's constellation of symptoms, the patient was evaluated for CRPS and felt to have met the criteria for this disorder based on the clinical Budapest criteria. The patient underwent 3 stellate ganglion blocks with a time interval of a week utilizing an anterior approach with fluoroscopic guidance. At each block, the patient received 3 mL of 0.25% bupivacaine and 2 mL of 8 mg dexamethasone.

The effectiveness of the block was recorded by observing the Horner's syndrome response after the blocks and decreased pain scores after all 3 procedures. A decrease in pain score was noted postprocedure (preprocedure pain score Visual Analog Scale 9 to postprocedure 4). Decreased edema on the back of the hand was observed at the 24th day postprocedure (Fig. 1).

## Discussion

PHN is one of the most common complications of acute HZ infection resulting from reactivation of the varicella-zoster virus (13). The natural course of acute

HZ infection is a self-limiting unilateral, painful, dermatomal vesicular rash. PHN is defined as persistent pain associated with zoster that is still present one month after the development of vesicles with acute HZ infection, and this painful period may persist for 3 to 6 months.

The most common complication of HZ is PHN. The incidence of PHN increases with age, and it has been reported as 25% to 50% in people over 50 years of age (8). Less common neurological complications are peripheral motor paresis and central nervous system involvement (e.g., meningitis, encephalitis, myelitis, and cerebral angiitis) (9). HZ-related CRPS was known, but there are few reports about this in the literature.

CRPS is a clinical syndrome with burning, pain, hyperalgesia or allodynia, edema, and sudomotor and vasomotor symptoms in the distal extremity that develops after a triggering factor. According to the terminology determined at the meeting of the International Society for Pain Studies in 1995: CRPS type I (develops as a result of surgery, immobilization, and trauma without nerve damage) and CRPS type II (with nerve damage) (10). The pathophysiology of this disorder is not fully understood, but it is thought to be related to changes in the sympathetic nervous system and/or neurogenic inflammation (11). Abnormal impulse generation from damaged nerves is responsible for the development or chronicity of symptoms, particularly in CRPS II. Although the role of the sympathetic nervous system in the underlying mechanism of CRPS-like symptoms described in PHN is explained by  $\alpha$ -adrenergic receptors, the use and effectiveness of sympathetic nerve blocks are unclear. Russo et al (12) presented a 4-component model of tissue trauma, pathological pain processing, autonomic dysregulation, and immune dysfunction to explain the CRPS pathophysiology and its cardinal features.

The inflammation process in HZ suggests that C-fibers, primary afferent neuron adrenoceptors, prolonged inflammation through neurogenic inflammation, may lead to central sensitization and may lead to CRPS-like symptoms with the contribution of immune-autoimmune processes added to the process. Not damage to C-fibers alone, but sustained C-fiber stimulation of a subacute-chronic inflammatory process and inappropriate interaction of nociceptive-sympathetic fibers may explain this process. It should be kept in mind that such CRPS-like symptoms may be seen after HZ infection, which is closely related to nervous system involvement and can lead to chronic inflammation, although not for all infections.



Fig. 1. (a) C5-C6 dermatomal rashes extending to the distal right upper extremity after HZ infection. Edema on the dorsum of the right hand, trophic changes in the nails, dermatomal rash scars are seen in the patient who developed CRPS after HZ infection. (b) After the right stellate ganglion block at the 24th day, a significant decrease in edema is shown in the dorsum of the hand.

HZ, herpes zoster; CRPS, complex regional pain syndrome.

The occurrence of CRPS-like symptoms during and after HZ is clinically known, but only 13 cases have been reported to date and nothing is known about the incidence or prevalence (6). When the literature was reviewed for patients with CRPS-like symptoms after HZ, early invasive interventions (e.g., recurrent stellate ganglion block, continuous epidural infusion, intravenous ketamine infusion, etc.) were performed in 3 patients and the symptoms were completely resolved (13-15). Conservative-medical treatments were applied in 7 patients, and symptoms resolved in 2 patients. (16-20). In the management of this case, we achieved positive results by using interventional methods in the early period in our patient who did not respond to medical treatment. Early interventional methods should always be kept in mind in HZ patients who develop CRPS after HZ or who progress with CRPS-like symptoms.

One study (6) showed that CRPS-like symptoms developing in HZ patients were only found in patients with a herpetic rash that spread to the affected extremity. In addition, in the same study, it was concluded that the more distal a herpetic rash on an extremity, the more severe and prolonged the clinical course of CRPS-like symptoms, and the greater the risk of progression to PHN (6). Based on this observation, predictive more aggressive treatment options and early application of

interventional procedures may limit the development of PHN in these cases.

Our patient had diffuse dermatomal eruptions distal and proximal to the upper extremity (i.e., arm, forearm, and hand), and prominent CRPS-like symptoms in the distal. We can associate the good prognosis with the early interventional treatment process in our patient who had pain control after sympathetic block and regression in edema.

## CONCLUSIONS

The occurrence of CRPS-like symptoms in patients with HZ infection is rarely, especially in patients with HZ infection, affecting the distal extremity. It is unclear whether the CRPS-like symptoms shown in this case are similar to CRPS from other underlying causes or are specific for HZ.

In these cases, it is important to consider that different neuropathic pain syndromes may coexist. Early use of interventional techniques should be considered in the management of treatment in these complex situations. Prompt diagnosis and appropriate interventions are crucial to the management of these patients to prevent persistent pain. Prospective randomized controlled studies are needed for appropriate patient selection and timing of treatment.

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