

## Case Report

# A Case of Zoster Sine Herpete of the Trigeminal Nerve

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## Abstract

Varicella-zoster virus reactivation causes zoster (shingles), a syndrome characterized by severe pain and a vesicular rash. The present report details a case of varicella-zoster virus reactivation of the maxillary and mandibular division of the right trigeminal nerve without evidence of vesicular rash (zoster sine herpete). It is difficult to identify owing to no typical clinical signs such as vesicular eruption. Zoster sine herpete of the trigeminal nerve, in particular, is rarely reported. In this case, the diagnosis was based on clinical findings and was supported by the demonstration of an immunoglobulin G antibody. Zoster sine herpete of the trigeminal nerve, in particular, should be considered in patients with severe facial pain over specific dermatomes, if they do not demonstrate appreciable findings of traumatic neuropathy, tumor or herpes zoster.

Key words: Zoster sine herpete—Trigeminal nerve—Varicella-zoster virus—Neuropathy—Diagnosis

## Introduction

Reactivation of a varicella-zoster virus (VZV) in a dorsal root or Gasser ganglion may result in vesicular eruption and pain due to subsequent infection of the epithelium overlying the dermatome where the virus had remained latent. This entity is often accompanied by deep and severe pain, allodynia or dysesthesia. In general, such clinical symptoms make diagnosis of VZV infection relatively easy. Clinically, we may also encounter zoster sine

herpete (ZSH)<sup>8)</sup>, which is more difficult to identify owing to no typical clinical signs such as vesicular eruption. Few articles, however, have been published on ZSH of the trigeminal nerve<sup>1,3)</sup>. Here, we report a case of ZSH of the trigeminal nerve treated at our institute.

## Case

The patient was a 48-year-old woman who visited Tokyo Dental College Suidobashi



Fig. 1



Fig. 2



Fig. 3

No clear findings corresponding to the symptoms encountered were observed on panoramic (Fig. 1), antero-lateral (Fig. 2), or Waters (Fig. 3) images.

Hospital. She complained of an abnormal sensation and pain in an area innervated by the right maxillary and mandibular nerves, which included teeth, oral mucous membrane and facial skin. Three months prior to this, she had experienced a dull, constricting pain, an itching sensation and hypoesthesia in the same region. She subsequently visited a private dental clinic where she was given occlusal treatment involving use of a bite splint. The intensity and frequency of both the abnormal sensation and pain had gradually increased, however. Her dentist was unable to make a diagnosis based on the symptoms

presented with. One month before attending our clinic, MR imaging and CT were performed on her brain and related structures at a department of neurology in a general hospital, but no abnormal findings were identified on the images obtained. The attending physician at the hospital prescribed carbamazepine. The symptoms showed no decrease, so the medication was terminated to avoid potential side effects such as nausea and dizziness. Imipramine also proved ineffective, and prednisolone only resulted in a slight reduction in pain.

On visiting our hospital, clinical examina-



Fig. 4 An increased signal area was observed in the right maxillary sinus indicative of mucous cyst, but no appreciable appearance accordant with the symptoms encountered on T2-weighted MR images.

tion identified abnormal sensation and pain. A continual and dull, constricting pain was present in the right upper and lower teeth. The patient also complained of no sensation or gustation in the tongue. A dull, constricting, background pain, dysesthesia and intermittent stabbing pains were also noted in the area innervated by the infraorbital and mental nerves. No other abnormalities were found in cutaneous or mucocutaneous areas or cervical lymph nodes.

A blood examination revealed that the total white cell count was 4,530 cells/ $\mu$ l, neutrophilia was 68.3%, lymphocytes were 24.9% and C-reactive protein was 0.0 mg/dl. These results indicated that there were no inflammatory changes. Radiography also revealed no dental, osseous or bone marrow abnormalities in the maxillo-facial region (Figs. 1–3). Magnetic resonance imaging was performed at another medical institution at our request to establish whether the abnormal sensation and pain were caused by a tumor. The results revealed an increased intensity mass in the right maxillary sinus on T2-weighted images (Fig. 4). It was diagnosed as a mucous cyst,

suggestive of no appreciable finding. She was aware of this lesion before her present symptoms had appeared 3 months earlier. No abnormality suggestive of abnormal sensation or pain was identified on T2-weighted MR images.

The sensory threshold in the right facial area was assessed by the Semmes-Weinstein test using a von Frey filament. The results showed 2.83 on the upper lip, 3.84 on the lower lip, 2.36 at the infraorbital foramen, and 4.08 at the mental foramen, whereas the normal range for the mechanical touch threshold is between 1.65 and 2.36. This indicated hypoesthesia in the area innervated by the infraorbital and mental nerves. Anti-VZV antibody values were measured by enzyme-linked immunosorbent assay. The results for VZV-immunoglobulin M (VZV-IgM) were negative. The titer for VZV-immunoglobulin G (VZV-IgG), however, was 1:51.3. These results suggested reactivation of VZV. Furthermore, no vesicular eruption in association with a specific dermatome was observed. Imaging findings showed no evidence of tumor or inflammation indicative of abnormal sensation or pain over the area concerned. From these results, we diagnosed this entity as ZSH of the trigeminal nerve.

The intensity of the pain was assessed by the visual analogue scale (VAS). The VAS value was 60 mm. The pain was severe enough to cause sleep disturbance. There was also potential masticatory disorder. Her pain relief score decreased from 10 to 0 after application of Stellate ganglion block (SGB), which was performed 13 times. She was prescribed amitriptyline hydrochloride at 30 mg/day and the pain gradually decreased. Thirty days later, her VAS score decreased to 30 mm. At that time, the results of the Semmes-Weinstein test showed 1.65 on the upper lip, 3.22 on the lower lip, 1.65 at the infraorbital foramen and 4.08 at the mental foramen. The hypoesthesia appeared to improve. A 70% improvement in gustation was also noted. Although she continued to receive treatment, no further improvement in her symptoms was observed.

Table 1 Different types of infection with varicella-zoster virus (VZV)

	Primary infection with VZV (chickenpox)	Reactivation of VZV (herpes zoster)	Reactivation of VZV (zoster sine herpette)
Type of infection	Droplet contact	Latent infection	Latent infection
Patient's condition	Fever and weariness	No particular	No particular
Distribution of vesicular rash	On body	Corresponds to specific dermatome	None
Neuralgia/abnormal sensation	Itching	+ / +	+ / +
IgM/IgG	+ / -	+ / +	+ / +

## Discussion

A patient presented at our hospital complaining of abnormal sensation and pain in an area innervated by the right maxillary and mandibular nerves. A lesion was identified and diagnosed as ZSH caused by VZV. The difficulty in diagnosing ZSH led to a delay in commencement of treatment.

The primary sign of VZV infection is the appearance of a chickenpox rash. This virus usually remains latent, however, in multiple dorsal root ganglia or Gasser's ganglia for an extended period of time. In immunocompetent elderly persons or immunocompromised patients, however, VZV frequently reactivates a few decades after initial infection to produce shingles (zoster). Reactivation of VZV leads to infection of the epithelium, a syndrome characterized by pain and the appearance of cutaneous or mucocutaneous vesicular eruption in not more than three dermatomes<sup>2</sup>. In a case of ZSH, the appearance of vesicular eruption may be inhibited by immunoreaction. Therefore, pain and an abnormal sensation associated with a particular dermatome are the main symptoms<sup>6</sup> (Table 1). To obtain an early diagnosis of ZSH, not only serologic examination, but also polymerase chain reaction (PCR) technology is important. It has been reported that PCR assay was used to detect VZV-DNA in saliva and determine reactivation of VZV<sup>4,7</sup>. In this case, the patient consulted us 3 months after recognizing her primary symptoms. Therefore, the chance of

an earlier diagnosis of ZSH by means of PCR assay was missed and treatment by administration of anti-viral drugs or steroids and SGB delayed. Stellate ganglion block and amitriptyline hydrochloride worked well for stubborn pain in this lesion. However, the pain did not disappear completely.

The type of pain resulting from a herpes zoster has variously been described as severe and deep, or as a boring, stabbing, aching, burning, pricking, tingling, or itching sensation. Such pain is intolerable, making control important, and is divided into 3 types according to the stage of vesicular eruption: preherpetic neuralgia, acute zoster pain and postherpetic neuralgia<sup>5</sup>. It is difficult to differentiate preherpetic neuralgia from postherpetic neuralgia by clinical findings alone as ZSH does not indicate vesicular eruption. Therefore, a clinical history and/or findings from serologic examination are necessary to make an appropriate diagnosis. Management for herpes zoster differs from that for postherpetic neuralgia. For preherpetic neuralgia, acyclovir or valaciclovir hydrochloride is used, but the treatment for postherpetic neuralgia is directed at managing the signs and symptoms. The treatment of ZSH is same as that for herpes zoster. In treatment of ZSH, we have to specify which type of pain the patient has. In this case, the VZV-IgM was negative, and 3 months before the patient had experienced an abnormal sensation and hypoesthesia in the area innervated by the right maxillary and mandibular nerves. Based

on the medical history of the patient and the outcome obtained, we diagnosed this lesion as postherpetic neuralgia caused by ZSH.

In conclusion, zoster sine herpete of the trigeminal nerve should be considered in patients with severe facial pain over specific dermatomes, if they do not demonstrate appreciable findings of traumatic neuropathy, tumor or herpes zoster.

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