Maxillary Alveolar Bone Necrosis Associated With Herpes Zoster Along Maxillary Nerve. A Rare Case Report with Review

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ABSTRACT
Herpes zoster is a viral infection caused by the reactivation of the varicella zoster virus, an infection most commonly affecting the thoracolumbar trunk. Herpes Zoster Infection (HZI) may affect the cranial nerves, most frequently the trigeminal. HZI of the trigeminal nerve distribution network manifests as multiple, painful vesicular eruptions of the skin and mucosa which are innervated by the infected nerves, usually associated with severe pain. Oral manifestations of HZ appear when the mandibular or maxillary divisions of the trigeminal nerve are affected. Reports of osteonecrosis and spontaneous tooth loss following herpes zoster infection of the fifth cranial are extremely rare. We are reporting a case of Herpes zoster involving the left side maxillary nerve with osteonecrosis of the maxilla.

Keywords: Maxilla, Osteonecrosis, Tooth exfoliation, Varicella zoster

INTRODUCTION
The varicella zoster virus (VZV) produces two clinical results: varicella or chickenpox and herpes zoster infection (HZI)¹. Varicella caused by the primary infection of VZV is a benign childhood disease producing eruptive vesicles. As a result of primary infection in the varicella, a skin virus moves to a sensory nerve and remains in latent state in a ganglion¹². When VZV in latent state is reactivated, it develops into HZI, which causes severe pain and painful vesicles in the skin and mucosa around the affected sensory nerve distribution³⁴.

Thoracolumbar dermatomes (T3-L3) are most commonly affected by HZI¹⁴. HZI may also affect the cranial nerves, with the trigeminal nerve most frequently affected (18.5-22%)¹. Herpes zoster affecting the trigeminal nerve is generally unilateral; it affects a single branch among three branches, mainly the first branch or optic nerve.
Oral manifestation can be observed when the maxillary and mandibular branches are affected\(^4\).

Oral vesicles appear mainly after a skin manifestation\(^3,5\). Sometimes, however, there may be mucosal involvement without skin lesion\(^5\). The vesicles erupt and leave mucosal erosions but no scar in most cases. The most significant complication of HZI is post-herpetic neuralgia\(^5\); other complications may include facial scarring, motor nerve palsy, optic neuropathy, blindness, encephalitis, and calcinosis cutis\(^6\).

The bony change in association with HZI was first reported by Rose in 19087. According to Dechaume et al. (1955)\(^8\), Gonnet’s presentation in 1922 was the first report to establish interest in osteonecrosis and tooth exfoliation associated with HZI\(^2\). Complication such as osteonecrosis with spontaneous tooth exfoliation is very rare. Thus, we report cases of osteomyelitis in the left side maxilla caused by HZI affecting the trigeminal nerve.

**CASE REPORT**

A 65-year-old male presented to the Our Department with complaints of pain of left side anterior maxillary region. On examination extra orally scarring and pigmentation was seen on left cheek region just below the lower eye lid spreading upto the ala of the left side nose and commissure of the lip fig 1. On intra oral examination exposed edentulous alveolar bone with respect to left side canine and premolar (fig - 2) was seen. On enquiry patient gave history of severe pain along with vesicular eruptions over the left cheek for 12 days. Eruptions were noted in successive crops over the left cheek, side of the nose, upper lip. Eruptions were heralded by mild fever, malaise, and piercing pain over the left cheek for initial two days. The vesicles were followed by pain and difficulty in chewing.

The patient was treated with oral acyclovir 800 mg. five times a day for seven days, supplemented by multivitamins. Nonsteroid anti-inflammatory analgesics were not effective in resolving in pain, so carbamazepine 200mg and Gabapentin were used to control intractable pain and itching, along with local application of topical anesthetics intraorally and calamine lotion extraorally.

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**Fig 1.** Scarring left side nose and facial Pigmentation

**Fig 2.** Exposed alveolar bone.
DISCUSSION

The prevalence rate of HZI in all ages is reported to be 1.2-4.8 per 1,000 people every year, with 7.2-11.8 people over the age of 609. The prevalence rate and seriousness increase with age. 40-50% of the patients with HZI are over the age of 60 every year, with 50% of persons over 85 years old recording prevalence rate of at least once10. As to the reason for such prevalence rate increase, natural immunity decline according to age increase may be considered; the decline in VZV-specific cellular immunity according to age increase is supported1,10. The prevalence rate also increases among immune compromised patients such as patients infected with human immunodeficiency virus, hematologic malignant disease, and immune-mediated disorder and organ transplant patients, and the risk of HZI for such immune-suppressed patients also increases according to age1,9,10. Other risk elements may include external damage of the affected dermatomes, psychological stress, and race9.

The thoracic dermatome is most commonly affected, accounting for 50% of the total cases9. The cranial nerve may also be affected, with the trigeminal nerve most commonly affected (18.5-22% of the total cases) followed by glossopharyngeal nerve and hypoglossal nerve. In case of trigeminal nerve involvement, it is unilaterally limited to a single branch, mainly affecting the optic nerve2.

Oral symptom is observed when the trigeminal maxillary and mandibular branches are affected and skin lesion is mainly preceded, but a case starting with paresthesia of mental nerve was also reported11. The erythematous vesicle is developed in the oral cavity; it ruptures, forms an ulcer, and gets covered by white pseudomembrane2,5,11. Lymphadenopathy may appear in the submandibular area5. In addition, patients may complain of symptom similar to acute pulpits of the affected tooth and toothache12; root resorption and periapical lesion may also occur13. Since histological findings that are not significantly different from the osteomyelitis pattern show necrotic bone and inflammatory cell infiltration, the diagnosis of the relationship with herpes zoster can hardly be confirmed only by such histological findings; the diagnosis should take into account the clinical findings3,4,14. Among the findings, osteonecrosis of the jaw and natural tooth exfoliation are very rare complications4,5,15. Jain et al.4 conducted a review of literature on 41 cases of osteonecrosis of the jaw caused by HZI. The onset age range was from 6 to 85, with 8 cases of patients under the age of 40,10 cases of those between 40 and 60, and 12 cases of those over 60. The prevalence rate increased according to age, with no difference by gender4,16. It mainly appeared unilaterally in the maxilla or mandible of the affected skin. 13 patients had it in the maxilla, and 18 patients, in the mandible. There were 31 lost teeth in the maxilla and 44 teeth in the mandible, making the mandible a predilection site. Since the anterior teeth numbered 64 and the posterior teeth were 61, there was no significant difference in the anteroposterior position. The exfoliated teeth per patient were 0-7; in five cases, all the teeth of the affected quadrant were lost4. With regard to the time interval between the outbreak of HZI and osteonecrosis accompanied by tooth loss, Mintz and Anavi3 presented in 1992.
a report on the interval of an average of 21.2 days together with the occurrence of natural tooth exfoliation about 2-6 weeks after HZI manifestation through a review of 14 studies. Several authors\textsuperscript{5,11} reported that it occurred between two weeks after early infection with complication of osteonecrosis and tooth exfoliation, but other authors \textsuperscript{4} reported that it occurred between 3-12 weeks after HZI manifestation as a late complication. In the first case reported in this paper, the extraction of the mandibular right canine was performed about 4 weeks after HZI manifestation even though the mandibular first and second premolars were ruled out because those premolars were lost four weeks before the outbreak of skin lesion. In the second case, natural tooth exfoliation occurred about three weeks after HZI manifestation. The pathophysiological mechanism of osteomyelitis of the jaw caused by trigeminal herpes zoster is controversial, and several hypotheses are under discussion. The first hypothesis is that local vasculitis caused by the direct extension of neural inflammatory response to the adjacent blood vessels. This eventually may cause an infarction of the trigeminal vessels that accompany the trigeminal nerves supplying the jaws and cause bone necrosis by triggering ischemia\textsuperscript{16}. The second hypothesis is that the generalized infection of terminal nerves supplying the periosteum and periodontium is believed to cause vasculitis of the periosteum and periodontium and avascular necrosis over a large area\textsuperscript{5,15,17}. According to Gilden et al.\textsuperscript{18}, since VZV can also be invaginated to the vascular endothelial cell in the peripheral nervous system instead of being limited to the brain or spinal cord, it causes small ischemic lesion, possibly developing into necrosis and demyelination. The report can serve as grounds for the two hypotheses above\textsuperscript{18}. The third hypothesis can be a denervation of bone, but denervation only can hardly be thought to cause serious bone damage in a short time\textsuperscript{15}. The fourth hypothesis is that systemic viral infection renders damage to an odontoblast and brings about tissue denaturation, causing pulpal necrosis. The final hypothesis is that the existing pulps, periodontitis, or surgical procedure around the HZI area can cause serious necrosis of the alveolar bone\textsuperscript{3}.

The literature review reveals that, in case of the outbreak of trigeminal herpes zoster, the prompt application of antiviral agents, active use of painkillers, and effective regulation of topical factors will be helpful in preventing such complications\textsuperscript{14,9,10}. If osteomyelitis and osteonecrosis of the jaw occur as complications, such complications can be treated through the proper use of antibiotics to avoid secondary infection, squestrectomy, removal of inflammatory tissue, and regular follow up\textsuperscript{2,4,13,15}. In cases, when osteomyelitis of the jaw and osteonecrosis accompanied by tooth exfoliation as rare complications after the outbreak of trigeminal herpes zoster occur, active use of painkillers, regulation of topical factors, and proper extraction of dead bone and affected teeth bring about good results.\textsuperscript{19}


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