CORRESPONDENCE



Involvement of the spinal trigeminal nucleus secondary to herpes zoster in a patient with hemifacial redness and swelling

Reactivation of varicella-zoster virus (VZV) in the trigeminal ganglion is related to immunity, often evidenced by the appearance of facial blisters. However, in a small number of patients, hemifacial swelling develops after the blisters have disappeared.¹ Here, we report the case of a patient with herpes zoster (HZ) characterized by marked hemifacial swelling and hyperintensity of the spinal trigeminal nucleus (SpV), as revealed by magnetic resonance imaging (MRI).

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One month before her first visit to our hospital, a 73-year-old woman developed pain on the right side of her head. She had no history of an immunocompromised condition (diabetes or malignancy) and had not received a COVID vaccine. One week later, blisters with a red halo appeared in the involved area, and on the right side of her forehead, together with swelling of the right eyelid. She was diagnosed with HZ by a local dermatologist. Her blisters and eyelid swelling disappeared after oral treatment with amenamevir. However, 3 weeks after the onset of pain, swelling of the right eyelid recurred.

During the first hospital visit, post-HZ pigmentation extended from the right forehead to upper eyelid. Also, redness and swelling were present on her right forehead, eyelids, cheek, lips, and right oral mucosa. The involved areas coincided with the area of branches 1–3 of the trigeminal nerve (Figure 1A–C). Her cerebrospinal fluid was positive for anti-VZV IgG (12.8), with elevated levels of neopterin (117.35 pmol/mL; reference: 30), white blood cells (17/µL; reference: 5.0), protein (35 mg/dL; reference: 15.0–45.0), and glucose (67 mg/dL; reference: 50.0–75.0). MRI showed a continuous region of hyperintensity extending from the right side of the trigeminal ganglion to the pons and medulla oblongata, consistent with the SpV



FIGURE 1 Photographs of the front side of the patient's face (A), upper lip (B), and right edge of the tongue (C) on the day of admission. She had multiple pigmentations in the area from the right forehead to upper eyelid (arrow), and redness and swelling of the right forehead, eyelids, cheeks, and lips. Erosions on the right edge of the tongue also developed. Her cutaneous and mucosal disorders were consistent with the involvement of the area served by branches 1–3 of the trigeminal nerve. On day 2 of hospitalization, T2-weighted magnetic resonance images revealed a continuous region of hyperintensity (arrow) extending from the right side of the trigeminal ganglion (D) to the pons (E) and medulla oblongata (F), consistent with the involvement of the spinal trigeminal nucleus. On day 3 of hospitalization, she developed severe redness and swelling of the right face (G)

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(Figure 1D–F). She was diagnosed with an SpV disorder secondary to HZ. As her symptoms worsened even after acyclovir infusion (750 mg/day) (Figure 1G), she was switched to combination therapy consisting of an infusion of 1500 mg acyclovir/day and 1000 mg methylprednisolone/day. The hemifacial swelling and redness of her face resolved, as did the hyperintense region on MRI.

The SpV is located outside the brainstem and is the sensory tract of branches 1-3 of the trigeminal nerve. Haanpää reported that 9 of 16 patients with HZ in the trigeminal and cervical nerve regions showed areas of hyperintensity in the brainstem and medulla oblongata on MRI.² In our patient, VZV may have been centrally transmitted from the trigeminal ganglion to the SpV.³ However, the disorders or symptoms that occur when VZV is transmitted to the SpV are largely unpredictable. Autonomic nerve neurons run in the SpV and are involved in autonomic nerve reflexes. Stimulation of the SpV can cause vasodilation of the facial skin.⁴ Because the roles played by the SpV differ from the roles of the principal sensory trigeminal nucleus and trigeminal motor nucleus, our patient exhibited no tactile disturbance or masticatory movement disorder. In addition to our patient, two cases of hemifacial swelling after HZ, manifesting as hyperintensity of the SpV on MRI, have been reported.^{1,5} Both cases required intravenous acyclovir and methylprednisolone pulse therapy.

In conclusion, we presented a rare case of hemifacial swelling and redness after an eruption of HZ. The pathomechanism likely involved stimulation of the SpV by VZV. MRI allowed early detection of the lesion.

DECLARATIONS

Approval of the research protocol: N/A.

Informed Consent: Informed consent was obtained from the patient. Registry and the Registration No. of the study/trial: N/A. Animal Studies: N/A.

KEYWORDS

herpes zoster, magnetic resonance imaging, spinal trigeminal nucleus, trigeminal nerve, varicella-zoster virus

CONFLICT OF INTEREST

The authors declare no conflict of interest.

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