**Varicella-zoster Virus Infection and Nummular Headache: A Possible Association with Epicranial Neuralgia**

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**Abstract**

A nummular headache (NH) is a type of primary headache that results from cranial neuralgia without a known cause. We herein report the case of a woman who suffered two episodes of focal headache in the left parietal area with identical characteristics that were compatible with NH. During the recovery phase of the second NH episode, the pain resurfaced with shingles coinciding with the painful area. The patient's NH subsided in parallel with resolution of the shingles. These findings support a diagnosis of peripheral neuralgia with NH. Latent virus infections, such as Varicella-zoster virus, that frequently cause distal nerve damage in patients with zoster sine herpete may be associated with epicranial neuralgia and NH.

**Key words:** nummular headache, varicella-zoster virus, shingle, zoster sine herpete, neuralgia, neuropathy

**Introduction**

The phenomenon of nummular headache (NH), or coin-shaped cephalgia, was first reported in 2002 (1). The incidence of NH is estimated to be 6.4/100,000/year (2). NH differs from other headache syndromes in its characteristics of consistent location, size and shape of the affected area during each attack (3). In the appendix of the second edition of the International Classification of Headache Disorders (ICHD-II), NH was categorized as a component of “cranial neuralgia and central causes of pain” (3). Despite reports of a few cases of intracranial secondaries (4-6), the etiopathogenesis of NH is factually unknown at present. We herein report the case of a woman who subsequently developed shingles in an area affected by NH. A seropositivity of IgM and IgG to Varicella-zoster virus (VZV) was identified. The relationship between viral neuralgia of latent viruses and NH is discussed.

**Case Report**

A 62-year-old woman suffered an acute onset of first headache 18 months ago. Her pain was confined to the left high parietal area in an oval shape with a clear margin. The area measured approximately 4.0×3.0 cm. The pain was sharp and electric in nature and was described as being located just beneath the surface of the skin. It occurred spontaneously, reached its maximum point within 10 minutes, persisted for hours and then later subsided. The headache was continuous with episodic exacerbation. Neither aura nor any associated features were reported. The intensity of the pain was mild to moderate. Triggering, aggravating or relieving factors were denied; however, the pain was reported to significantly worsen when brushing her hair or pressing the scalp in the painful area (static and dynamic allodynia) during an attack of pain. The patient denied any antecedent cutaneous vesicle formation, craniofacial trauma, recent infection, chemical exposure or previous history of migraine, stroke, herpetic labialis or shingles. She had been diagnosed with chronic inflammatory demyelinating polyneuropathy three years prior to the attack. Diagnoses of lymphoma and IgM macroglobulinemia were later confirmed, and both remitted after the patient received appropriate treatment.

The patient’s first visit occurred two days after headache onset. No papilloedema was observed in the fundus. A neurological examination showed no consistent abnormal findings of higher cortical function, cranial nerves, motor function, sensory system, equilibrium or co-ordination. No percussion pain at the sinus or spine or nodulation or tender-
The patient was referred to our service 10 days after recurrence. Upon presentation, clusters of vesicles with erythematous bases were seen in the affected area and in the left ear (Figure). A dermatologist confirmed a diagnosis of shingles based on the cutaneous findings and a seropositivity of IgM-VZV and IgG-VZV. Famciclovir was administrated. Head magnetic resonance imaging and autoimmune indices did not disclose any consistent abnormalities. The dose of gabapentin was then increased to 300 mg/day. The patient’s pain completely subsided within six weeks, and a seronegativity of IgM-VZV and scarring of the scalp were noted at that time (Figure). Gabapentin was then discontinued. The patient’s lymphoma and macroglobulinemia did not recur during the course of treatment.

Discussion

For our patient, the occurrence of shingles in an area affected by recurrent headaches may be argued to be an accidental coincidence. However, the congruity of coverage, the characteristics of pain and the simultaneous recovery between the shingles and NH reasonably support the presence of an association between shingles and NH. Therefore, the findings of our single patient clearly provide two rational pieces of evidence regarding NH: a peripheral origin of pain and a probable infection with a latent virus responsible for neuralgia.

Until now, there have been two theories postulated regarding NH. The central theory proposes a central sensitization of a pain pathway in NH. This theory is supported by a few cases of intracranial secondaries associated with NH (4-6), the complete recovery of pain in a patient after the removal of a meningioma (6) and the failure of pain control in nerve block. On the other hand, the peripheral theory proposes the involvement of nociceptive receptors or terminal endings in local epicranial tissue. This theory is broadly encouraged by the consistent characteristics of pain at the affected area during each pain attack, as well as the alteration of focal sensory algometry (7) or trophic changes (8) confined to the affected area in some NH patients. The superficial location of pain and stimulus-evoked (static and dynamic mechanical stimulus) allodynia provide additional evidence for a peripheral origin of pain in NH. Accordingly, the clinical findings in our patient clearly support a peripheral origin of NH pain.

Besides the mention of antecedent head injury in a few reports of NH (1), the cause of neuropathic change in NH has not been identified; however, epicranial neuralgia is considered to be responsible for NH. In our patient, two episodes of painful attack and shingles occurred during the second episode of headache. Since the extent and characteristics of the pain did not change when the shingles occurred, we believe that the second episode of NH was compatible
with VZV-related neuralgia. In fact, VZV can cause neuropathic pain with cutaneous vesicle formation (shingles) or without skin rash (zoster sine herpete) in both the preherpetic, acute and postherpetic stages (9, 10). Importantly, post-herpetic neuralgia also occurs with a consistent location, shape and size (11), similar to that observed in our patient. Therefore, epicranial neuralgia in NH may be a form of zoster sine herpete.

The limitation to our findings is that NH patients are expected to present with other medical disorders if VZV or other latent viruses are responsible for the neuralgic pain. However, no such studies have been conducted in NH patients to date. Nevertheless, the simultaneous occurrence of VZV and NH in our patient establishes a viral model of epicranial neuralgia for NH. Therefore, NH may not be a primary headache or a headache with a central cause. A detailed investigation between latent viral neuralgia and NH is warranted.

Clinically, NH exhibits a shorter duration of pain than that observed in zoster-related pain. Regarding trigeminal neuralgia, the pain in both NH and trigeminal neuralgia locates superficially and exhibits consistent characteristics. However, the pain in NH persists for hours and does not have a specific triggering factor as in trigeminal neuralgia. Accordingly, NH, zoster-related pain and trigeminal neuralgia may belong to separate, specific forms of trigeminal pain.

In conclusion, we herein report the case of a woman who developed shingles in an area affected by NH two weeks after the onset of the second episode of NH. This association raises the notion of a potential relationship between latent virus infection and neuralgic pain in NH patients. Therefore, we suggest the need to establish an appropriate test for NH to uncover any latent or subclinical disorders.

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References