Acyclovir Treatment in 2 Patients With Benign Trigeminal Sensory Neuropathy

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Idiopathic trigeminal sensory neuropathy (TSN) is characterized by transient sensory disturbances in the distribution of one or more branches of the trigeminal nerve. Pain is not a typical complaint. Although the symptoms may last from days to years, complete recovery is normal in half of the patients.1,2

A viral theory has been postulated as the cause of TSN.3-8 In support of this hypothesis, herpes simplex virus oral mucosal lesions have been reported in patients with trigeminal neuropathies,9 and 2 cases of recurrent idiopathic TSN have been described.10 This report describes 2 patients with facial numbness diagnosed as TSN who had associated ulcerated oral lesions and who were cured with acyclovir. These results favor a viral etiology and suggest the possibility of treating TSN with antiviral agents.

Report of Cases

The diagnostic criteria for TSN are as follows1: 1) sensory loss symptoms limited to the territory of one or more of the 3 branches of the trigeminal nerve; 2) absence of any underlying condition that could have explained the symptoms, especially dental and facial trauma, neoplasms, mul-
CASE 1
A 38-year-old woman presented with a history of episodic seasonal left facial numbness over the past 10 years. The symptoms reappeared each year in autumn, lasted the entire winter, and disappeared in the summer (ie, duration of 6 to 8 months). We had witnessed the seasonal occurrence of symptoms on 2 occasions.10 There were no other antecedents of disease. Numbness began at the tip of the tongue, gums, and lips and spread through the lower half of the face to affect the entire left side of the face (Fig 1). The patient had difficulty speaking and swallowing because of repeated inadvertent biting of the tongue and lips. The lips and tongue were always the last regions to improve.

Clinical examination revealed moderate hypoesthesia to pinprick and touch in the region of the second and third branches of the left trigeminal nerve. The remainder of the neurologic examination, which included corneal reflexes and trigeminal motor function, was normal. Intraoral examination revealed a small fibroma on the tip of the tongue, caused by biting, and a small ulceration of the gum adhered to the lower right first premolar. Urinalysis, blood tests, and the sedimentation rate were normal. Serology for syphilis was negative. Serum immunoglobulin (Ig) G antiherpes simplex virus antibodies (1:1,280) and serum IgG antiherpes zoster antibodies were positive (1:1,200 and 1:640, respectively). An enzyme-linked immunosorbent assay showed that the IgM antiviral antibodies were negative. A CT scan of the skull base and mandible were normal. A magnetic resonance imaging (MRI) study of the brain was also normal. The neurophysiologic study of the trigemino-facial reflex (blink reflex) and trigeminal somatosensory evoked potentials were normal on both sides of the face.

Oral acyclovir was given (1,000 mg daily, for 10 days) starting from the appearance of symptoms (ie, when perioral numbness was first noted). The symptoms disappeared entirely after the treatment. The patient was followed for 3 more years; the first year numbness did not develop in the usual winter period, whereas in the subsequent 2 years numbness did appear and again disappeared after receiving acyclovir treatment.

CASE 2
A 27-year-old man was referred with a history of the appearance of small ulcers in the oral cavity 2 months before, followed 1 week later by oral numbness similar to that experienced under dental anesthesia. The numbness began in the lower lip and spread from the oral commissure toward the region of the third branch of the right trigeminal nerve.

Examination revealed trigeminal hypoesthesia in the territory of the third branch of the right side (Fig 2). The remainder of the neurologic and oral examination was normal, as were urinalysis, blood tests, and sedimentation rate. The serum IgG antiherpes simplex virus antibodies titer was greater than 2,560, and no IgM antiviral antibodies were present. Oral acyclovir was given (1,000 mg daily for 10 days), with total disappearance of symptoms. Follow-up for 2 years showed no recurrence of symptoms.

Discussion
Facial numbness may only be considered idiopathic when other possible causes have been excluded and when a careful, prolonged patient follow-up is performed. A differential diagnosis that includes malignant disease must always be established in the case of facial numbness localized to the distribution of the third branch of the trigeminal nerve, or that starts in the region of the oral commissure and gradually spreads to the entire side of the face.11-13 Collagen disease must also be excluded.14,15

Patients with TSN commonly lack disease antecedents of importance or associated neurologic disorders. Trigeminal motor function is usually preserved. According to Fisher,6 pain is present in approximately one of every 3 cases in the initial stages of the disease. However, no associated pain was noted in 2 cases. Sensory deficiency varies from slight to severe,4,5,16,17 and was found to be moderate in these patients. In decreasing order of frequency, hypoesthesia affects all the trigeminal branches; the second and third branches; the third branch only; the second branch only; and the first and second branches. Thus, the
second and third trigeminal branches are affected with approximately the same frequency and twice as often as the first branch. In the present cases, one patient had involvement of all three branches, and the second suffered hypoesthesia in only the region of the third trigeminal branch.

Sensory loss is well defined and corresponds to the territory innervated by each of the affected trigeminal branches. This suggests that the underlying lesion in benign TSN could be located in the trigeminal ganglion or peripherally. However, the general absence of pain is an argument against ganglion involvement.

The true cause of this syndrome remains open to debate. Hughes observed arachnoiditis of the trigeminal sensory roots in 3 cases during surgery. However, the latter could not be confirmed, because the cytology and histopathology were nonspecific in the first case, whereas in the second case the lesions healed before the patient sought help for the numbness.

In both patients, the facial numbness disappeared after administration of acyclovir. These data further support the viral theory of spontaneous and transient trigeminal numbness and raise the question of whether patients with TSN should be treated with acyclovir.

References