Trigeminal Neuralgia Induced by Sour and Spicy Foods: What Is the Underlying Mechanism? A Case Report

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This article is a case report of a female patient in whom sour and spicy foods evoked trigeminal neuralgia (TN). An attempt to reveal the underlying pain mechanism is described and discussed. The 81-year-old woman had been suffering from classical TN since the age of 50. Attacks occurred spontaneously or in response to mechanical stimuli. In addition, sour and spicy foods also evoked TN attacks and were therefore avoided for years. Medical treatment was initially effective, but two radiofrequency ablations of the gasserian ganglion were required later on and yielded good, albeit incomplete, pain relief. Sensory examination consisted of application of a mechanical stimulus and sweet, salty, sour, and spicy solutions to the anterior part of the tongue and the mandibular mucosa on both sides. Mechanical stimuli were felt but produced no pain. When applied to the tongue, the tastes of all solutions were identified but produced no pain. When applied to the mucogingival line, none of the solutions was identified but the sour and the spicy solutions provoked TN immediately following their application. These findings suggest that in this patient, sour and spicy solutions may have evoked TN attacks by direct activation of trigeminal C-nociceptors, possibly via interactions with transient receptor potential vanilloid 1 receptors. *J Oral Facial Pain Headache 2016;30:267–270. doi: 10.11607/ofph.1590*

Keywords: *facial pain, gustatory, sensory-testing, trigeminal neuralgia, TRPV1 receptors*

Trigeminal neuralgia (TN) is a disorder characterized by recurrent, unilateral, brief, electric shock–like pains, abrupt in onset and in termination and limited to the distribution of one or more divisions of the trigeminal nerve.[1] Attacks may occur spontaneously or in response to innocuous stimuli, mainly mechanical, such as light touch, tooth brushing, or cold air.[2] TN attacks in response to eating and drinking are well known and are generally attributed to the mechanical or, when relevant, to the thermal (cold) aspects of these activities, but only scarce reports have shown associations between specific food flavors and TN. In one case report of a patient with idiopathic TN, pain was provoked by application of sucrose to the ipsilateral anterior two-thirds of the tongue. Application of other stimuli such as saline, citric acid, and water had only a minimal effect.[3] In another patient, electric-like, short, unilateral pain was also evoked by strong sweet stimulation and treated effectively with carbamazepine.[4] In both cases, convergence between the gustatory and trigeminal pathways was hypothesized to be the cause of pain. One additional study reported elevated salty, sweet, and olfactory thresholds in patients with idiopathic TN, although the sour and bitter thresholds were normal. However, similar changes in gustatory thresholds were also found in patients with burning mouth syndrome and with trigeminal postherpetic neuralgia. In that study, induction of pain by gustatory stimuli was not reported by any of those patients.[5] The following is a report of an unusual case of a woman with long-standing TN in whom pain was repeatedly triggered by sour and spicy foods. The possible underlying pain mechanism is also presented.

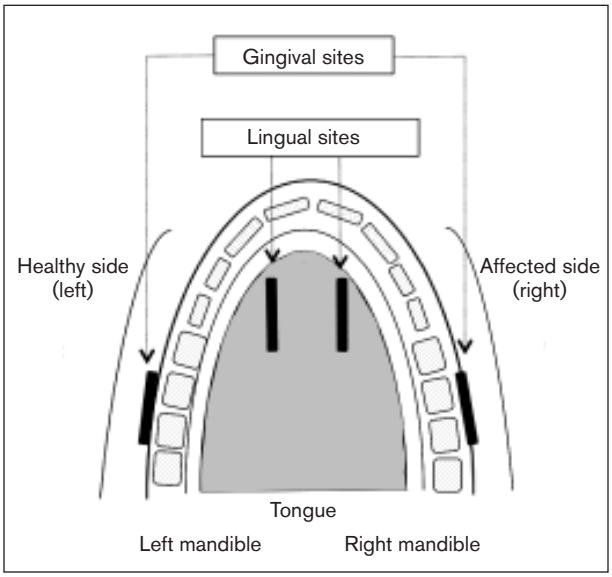

Fig 1 Illustration of the testing sites.

Case Report

An 81-year-old woman had been suffering from attacks of brief, stabbing pain in the right side of her cheek and chin since age 50. Pain occurred spontaneously and in response to light touch on the lower part of the face and to talking, chewing, and tooth brushing. In addition, she noticed that sour and spicy foods provoked especially intense TN attacks and she had therefore completely eliminated such foods from her diet over the past several years. This fact was left unnoticed by her treating physicians. The diagnosis of classical TN was made by a neurologist early on in the course of her illness and she had been successfully treated by carbamazepine for about 25 years. In 2008, at approximately age 75, the TN worsened without any clear reason (ie, no dental treatment, no trauma, etc) and limited her ability to talk and eat. A Magnetic Resonance Imaging scan (1.5 Tesla) revealed an unrelated old lacunar infarction in the right temporal region but no evidence of neurovascular compression or any other pathology in the cerebello-pontine angle or along the trigeminal nerve. The carbamazepine dose was increased to the level of tolerability and additional medications (baclofen, clonazepam, and gabapentin) were added. As pain persisted, a radiofrequency ablation of the right gasserian ganglion was performed in 2012 and again about 1 year later. About 2 months after the second procedure, the frequency and severity of the attacks declined and medication doses were gradually reduced.

Due to the unusual nature of the attacks of TN produced by foods, she was referred for a neurologic examination at the Institute of Pain Medicine at Rambam Health Care Campus in Haifa, Israel in July 2014. The findings from the general neurologic examination were unremarkable. Facial light touch and pinprick sensations were normal on both sides. No mechanical trigger zones were detected. The sensory examination of the oral cavity consisted of the following stimuli: First, three consecutive gentle strokes of a cotton swab were applied to both sides of the tongue and to the buccal aspect of the mandibular mucogingival line, just below the first and the second molars (gingival sites; Fig 1). The mechanical stimuli were felt but provoked no pain. Subsequently, sweet, salty, sour, and spicy solutions (made by adding a teaspoon of sugar, salt, vinegar, and ground chili pepper, respectively, to 100 mL of water) were applied to the two sides of the tongue, followed by testing the two gingival sites, with the healthy side always before the affected one. The sequence of the applied solutions was randomly selected so the patient could not know in advance which taste would come next. The patient was requested to indicate the perceived taste and whether or not it was painful by pointing with her finger to a board on which the four tastes as well as the indications "pain" and "no pain" were written. She was instructed to protrude her tongue and refrain from talking to avoid spreading the applied solutions to other regions of the mouth. The patient correctly identified all four taste solutions applied to both sides of the tongue, but identified none at the gingival sites. No pain was reported in response to the application of any of the four solutions to the tongue sites or to the unaffected gingival site. In the affected gingival site, both the sour and the spicy solutions evoked pain soon after their application. The pain was described as a few bursts of moderate to severe stabbing pain, which the patient recognized as "a typical TN attack." It was followed by a mild burning/aching sensation in the territory of the mandibular nerve in the affected side, which gradually subsided over several minutes. On the following day the patient confirmed that her pain remained under control as before.

Discussion

The exact pathophysiology of classical TN remains unclear, but it is widely accepted that vascular compression at the trigeminal root entry zone resulting in focal demyelination is the main precipitating factor.[6,7] The intraneural mechanisms which lead to the paroxysms typical of TN are also not entirely obvious. One explanation, the "ignition theory," suggests that axonopathy-induced changes in electrical excitability

such as ectopic impulse discharge, spontaneous and triggered afterdischarge, and cross excitation of neighboring afferents occur in trigeminal axons and neuronal cell bodies.[8] More recent studies suggest that the loss of central inhibition of the nociceptive system may also play a role in the pathogenesis of TN, especially in patients with background (ie, between paroxysms) pain.[9]

One of the main characteristics of TN is that the pain is provoked by nonpainful mechanical stimuli.[2] In contrast, other stimuli (eg, gustatory) rarely, if at all, produce pain in patients with TN. Only two case reports of TN evoked by gustatory stimuli can be found in the literature; sweet taste was the inciting stimulus in both of them.[3,4] In the first case, pain was evoked by applying 10% sucrose solution to the anterior two-thirds of the tongue ipsilateral to the painful side; pain was not evoked when applied to the contralateral side. In the second case, carbamazepine completely abolished "neuralgic-type pain," which was previously induced by placing half a teaspoon of sucrose on the patient's tongue.[4] In both cases, convergence between the gustatory and trigeminal pathways was hypothesized to be the cause of pain. Since no evidence for previous injuries of peripheral nerves was reported, the convergence between gustatory and trigeminal nociceptive afferent inputs has been suggested to occur at a central level. However, convergence between these two systems has been under debate; some studies have produced evidence supporting such convergence in both animals and humans[10,11] but a recent animal study suggested that gustatory nerve alterations do not affect trigeminal nociceptive transmission.[12]

In the patient presented here, the putative pain mechanism seems different. The administered solutions (salty, sweet, sour, and spicy) clearly activated the gustatory system since the patient correctly identified all four of them when they were applied to the tongue sites. However, the application of these solutions to the tongue did not evoke any pain, indicating that convergence between the gustatory and trigeminal nociceptive afferent inputs may not have been the pain-generating mechanism in this patient. However, application of the spicy and sour solutions to the affected gingival site did provoke pain, although the solutions were not identified by the patient. This suggests a pain-provoking mechanism that involves direct activation of trigeminal afferent inputs without involvement of the gustatory pathways.

The provocation of pain by applying the spicy solution to the patient may be somewhat different, since the spicy solution may not have necessarily involved the gustatory system (which is activated by five distinct tastes: sweet, sour, bitter, salty, and savory/umami[13]). Rather, spicy solutions may activate trigeminal nociceptive pathways.[14] However, since the spicy solution evoked pain only when applied to the affected gingival site and not to the tongue on the affected side, this finding suggests that pain occurred only in response to activation of the trigeminal nociceptive afferents within specific trigger zones, similar to what is commonly seen in mechanically induced TN.

From a chemical standpoint, the spicy and sour solutions consisted of capsaicin and vinegar (acid), respectively. Both capsaicin and acid have been shown to activate C-nociceptors by acting on the transient receptor potential vanilloid 1 (TRPV1) receptors, thus evoking pain.[15] TRPV1 receptors have been identified in nongustatory sensory afferent fibers, but evidence for their expression in gustatory afferent neurons is controversial.[16,17] Hence, if indeed correct, this observation suggests that in addition to the well-known involvement of myelinated afferents, unmyelinated C-fibers can also play a role in the pathogenesis of TN, since TRPV1 receptors are only located on C-fibers.[15] Also of relevance is a recent report of a 39-year-old woman who suffered from TN of the lingual nerve for 6 years, which was resistant to medical and surgical interventions. Daily mastication and consumption of very hot chili peppers resulted in complete resolution of her TN, suggesting again that the C-fibers may be involved in the pathogenesis of classical TN, at least in some patients.[18]

Several additional aspects of this unique patient deserve consideration. First, the patient has had two radiofrequency ablations in the past, which might have affected the results of the sensory examination presented here. However, the fact that sour and spicy foods caused TN attacks many years prior to these ablations and the fact that the sensory examination of the face was normal reduce the likelihood of this possibility. Second, although the burning pain which followed the bursts of the stabbing pain reported by the patient is not a typical feature of classical TN, background pain before or after attacks of TN have already been reported.[19]

Conclusions

The patient presented here showed a unique representation of TN that was induced by sour and spicy foods. The findings of a careful neurologic examination point to the potential involvement of TRPV1 receptors, and therefore possibly C-fiber afferents, in the pathogenesis of the patient's pain.

Acknowledgments

The author reports no conflicts of interest.

References

1. Headache Classification Committee of the International Headache Society (IHS). The International Classification of Headache Disorders, 3rd edition (beta version). Cephalalgia 2013;33:629–808.
2. Zakrzewska JM. Differential diagnosis of facial pain and guidelines for management. Br J Anaesth 2013;111:95–104.
3. Sharav Y, Benoliel R, Schnarch A, Greenberg L. Idiopathic trigeminal pain associated with gustatory stimuli. Pain 1991;44: 171–174.
4. Helcer M, Schnarch A, Benoliel R, Sharav Y. Trigeminal neuralgic-type pain and vascular-type headache due to gustatory stimulus. Headache 1998;38:129–131.
5. Siviero M, Teixeira MJ, de Siqueira JT, Siqueira SR. Somesthetic, gustatory, olfactory function and salivary flow in patients with neuropathic trigeminal pain. Oral Dis 2010;16:482–487.
6. Jannetta PJ. Arterial compression of the trigeminal nerve at the pons in patients with trigeminal neuralgia. J Neurosurg 1967;26(suppl 1):159–162.
7. Zakrzewska JM and Coakham HB. Microvascular decompression for trigeminal neuralgia: Update. Curr Opin Neurol 2012; 25:296–301.
8. Devor M, Amir R, Rappaport ZH. Pathophysiology of trigeminal neuralgia: The ignition hypothesis. Clin J Pain 2002;18:4–13.
9. Obermann M, Yoon MS, Ese D, et al. Impaired trigeminal nociceptive processing in patients with trigeminal neuralgia. Neurology 2007;69:835–841.
10. Nomura S, Mizuno N. Central distribution of afferent fibers in the intermediate nerve: A transganglionic HRP study in the cat. Neurosci Lett 1983;41:227–231.
11. Schöbel N, Kyereme J, Minovi A, Dazert S, Bartoshuk L, Hatt H. Sweet taste and chorda tympani transection alter capsaicin-induced lingual pain perception in adult human subjects. Physiol Behav 2012;107:368–373.
12. Boucher Y, Simons CT, Carstens MI, Carstens E. Effects of gustatory nerve transection and/or ovariectomy on oral capsaicin avoidance in rats. Pain 2014;155:814–820.
13. Iwata S, Yoshida R, Ninomiya Y. Taste transductions in taste receptor cells: basic tastes and moreover. Curr Pharm Des 2014;20:2684–2692.
14. Nilius B, Appendino G. Spices: The savory and beneficial science of pungency. Rev Physiol Biochem Pharmacol 2013; 164:1–76.
15. Willis WD Jr. The role of TRPV1 receptors in pain evoked by noxious thermal and chemical stimuli. Exp Brain Res 2009; 196:5–11.
16. Roper SD. TRPs in taste and chemesthesis. Handb Exp Pharmacol 2014;223:827–871.
17. Viana F. Chemosensory properties of the trigeminal system. ACS Chem Neurosci 2011;2:38–50.
18. Loeser J, Pilgram B, Dagtekin O. Chili for therapy of trigeminus neuralgia: a case report [article in German]. Schmerz 2012;26: 435–437.
19. Zakrzewska JM, Padfield D. The patient's journey through trigeminal neuralgia. Pain: Clinical Updates 2014;22:1–7.