Posttraumatic Gustatory Neuralgia: A Clinical Model of Trigeminal Neuropathic Pain

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Six cases are reported in which the primary complaint was episodic, recurrent facial pain that was triggered by a taste stimulus. The pain first occurred days to weeks after head and neck surgery. Patients reported that a food stimulus placed in the mouth evoked episodic, electric shock-like pain in a preauricular location on the surgical side. The smell of food or, less reliably, emotional excitement could also trigger pain. Mandibular movement did not evoke the pain, and between lancinating attacks there was either no pain or only mild discomfort. Following an episode of pain, there was a refractory period during which the pain could not be elicited. Physical examination demonstrated a preauricular sensory loss of variable distribution. No abnormal sweating or vasomotor findings were clinically apparent. No odontogenic, muscular, salivary gland, neurologic, or psychologic pathology was found to explain the clinical symptoms. The pain was not relieved with standard doses of anticonvulsants that are commonly used to treat trigeminal neuralgia. The duration of the recurrent pain symptoms in this group was 8 to 132 months without remission. Gustatory neuralgia may be a discrete syndrome that results from abnormal interactions between salivary efferent fibers and trigeminal sensory afferent fibers in the injured auriculotemporal nerve. The unique features of the disorder make it a potentially useful clinical model for the investigation of autonomic/sensory interactions in neuropathic pain.
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A distinct clinical syndrome that is associated with damage to the auriculotemporal nerve (ATN) has been recognized for over 200 years,[1–3] but it was not described as a separate entity until 1923 by Lucie Frey.[4] "Frey syndrome" is characterized by sweating and flushing in the cutaneous distribution of the ATN following a gustatory stimulus. Pain is an uncommon finding in this syndrome; pain occurs in less than 10% of cases, and it is usually described as constant aching or burning.[5,6] Reports associate this syndrome with surgical trauma (eg, parotid gland surgery), accidental trauma, local infection, viral illness, sympathetic dysfunction, central nervous system disorders, carotid endarterectomy, temporomandibular joint (TMJ) surgery, orthognathic surgery, oncologic surgery, combined cancer therapy, and idiopathic causes.[6–23]

In a small number of reported cases, pain was the dominant symptom,[6,22–24] and in three reports pain was triggered by gustatory stimuli.[22–24] Truax[22] described a case of paroxysmal, lancinating pain without facial sweating in the preauricular region that occurred only after gustatory stimuli following carotid endarterectomy. De Benedittis[24] reported two cases of gustatory-evoked pain; one was described as a case of deep, aching pain with no facial flushing or sweating, which lasted from 5 to 6 minutes and occurred without nerve injury, while the other case was "tic douloureux pain," or trigeminal neuralgia with flushing and sweating. Sharav et al[23] described a case of pain that lasted from 10 to 60 minutes, also without history of nerve injury. The clinical features of these few cases differ from the classic description of Frey syndrome, and they may represent a separate disorder with a different and unique pathophysiology.

This report details the clinical features of six cases in which only paroxysmal pain developed after surgery in the region of the ATN. In all six cases the pain was provoked by taste stimuli.

Materials and Methods

This multidisciplinary pain group evaluated patients that presented to the authors' pain center with a complaint of facial pain that was evoked by foods. All patients underwent a comprehensive interview and medical history, head and neck exam, dental evaluation, neurologic exam, and psychologic evaluation. The medical and dental workup included a routine physical examination, a cranial nerve exam, trigeminal neurosensory testing, and oral and dental exams.

The cranial nerves were clinically tested, with special attention paid to taste on the tongue and to the trigeminal system. A cotton-tipped applicator was used to apply salty, sweet, sour, and bitter flavors (no specific concentrations) to the tongue to test for response to specific tastes, and the responses were documented. Pain elicited by the application of substances to the tongue was documented and the area of pain was mapped. Additional sensory abnormalities in the trigeminal system were documented by means of response to sharp and light touch and hot and cold temperature, tactile contact discrimination that was assessed by means of von Frey hair stimulation, and two-point discrimination that was assessed with Bolger calipers; the results were then mapped. Evidence of autonomic dysfunction (gustatory sweating, lacrimation, and skin color changes) were documented and mapped. Parotid gland function was grossly evaluated by physical examination (stimulated and unstimulated), and any significant abnormalities were documented. Any evidence of dental and oral disease was also documented. Differential local anesthetic nerve blocks of the trigeminal system were performed and the responses were evaluated.

Patients received a diagnosis of "gustatory neuralgia" if they exhibited the following criteria: *(1)* paroxysmal, lancinating facial pain evoked by placing certain foods into the mouth, *(2)* pain only in the distribution of the ATN, *(3)* no present evidence of salivary gland pathology that would account for the pain, and *(4)* no evidence of dentoalveolar/odontogenic pathology that would account for the pain.

Results

A retrospective review of six patients that fit the clinical criteria for a diagnosis of "gustatory neuralgia" is presented in Table 1. All six patients experienced the onset of pain following a surgical procedure in the ipsilateral head and neck region; three had undergone TMJ surgery, one had parotid tumor surgery, one had oral cancer surgery, and one had a carotid endarterectomy. All patients reported a latency of at least 1 week or more before the development of pain. In all patients the pain was severe, electric-like, lancinating, and of short duration (seconds); the pain was located in the anatomic distribution of the auriculotemporal branch of the trigeminal nerve. The pain was evoked only by eating and by the application of certain foods to the ipsilateral tongue. The patients also complained of numbness in the preauricular region following surgery.

Clinical examination revealed a neurosensory deficit in the anatomic distribution of the trigeminal nerve without hypoesthesia, hyperalgesia, or allodynia. Upon tongue testing, the sensory examination was normal without mechanical hyperalgesia to light touch, sharp touch (pin prick), or temperature. The pain, however, could be elicited by the application of certain substances to the ipsilateral tongue (sour, sweet). Examination of the remainder of the oral and maxillofacial region was essentially normal.

All six patients had been given varied trials of several anticonvulsant and neuropathic pain medications that are typically used for trigeminal neuralgia-like pain, all without relief of the taste-evoked pain. Two patients had received lingual nerve local anesthetic block injections with sensory anesthesia and elimination of the taste-evoked pain. Two patients were considered for ablative neurosurgical procedures, but they refused treatment.

Table 1 Characteristics of Gustatory Neuralgia: Patient Profiles

Characteristic	Patient 1	2	3	4	5	6
Gender	Female	Male	Female	Female	Male	Female
Age at onset (y)	39	35	36	43	59	46
Duration (mo)	8	132	12	120	24	21
Delay of outset from surgery	Weeks–months	Weeks–months	Weeks	Days–weeks	Weeks	Months
Weight change	15-lb loss	None	20-lb loss	20-lb loss	15-lb loss	Minimal
Anterior/posterior of mouth	Anterior	Anterior	Anterior	Anterior	Anterior	Anterior
Side of face	Left	Left	Right	Right	Right	Left
Facial weakness	Ipsilateral postoperative	None	Ipsilateral postoperative	Ipsilateral postoperative	None	None
Symptom frequency	Every meal	Daily, when triggered	Every meal, "thousands" of times	Every meal	With triggers, every meal	With triggers, with eating at all times
Pain quality	Electric	Electric	Electric	Electric, lancinating	Electric, lancinating	Electric-like
Refractory period	Seconds	20–60 s	Seconds	Seconds	Seconds	Seconds
Pain intensity	5 out of 10	8 out of 10	Severe	Severe	10 out of 10	6 out of 10
Sensory triggers	Food, cola, cold liquids	Sour foods, citrus	Certain foods	Food in mouth	Toothpaste, lemon, citrus	Sour foods
Worst stimuli	Chocolate, lemon juice	Citrus, pineapple	Spicy foods	Spicy foods	Lemon	Lemon, spicy foods
Other triggers	Excitement, fear, smell of perfume	None	Sudden excitement	Sudden excitement	None	None
Abnormal sweating	No	No	No	No	No	No
Facial flushing	No	No	No	No	No	No
Past pain history	None	Facial pain, TMD	None	TMD	None	TMD
Headache history	Pregnancy migraine	Tension-type	None	TMD, tension-type	None	None
Medical history	TMJ surgery	TMD	None	Headache	HTN, DM, CAD, stroke	None
Current medical problems	None	None	None	Headache	HTN, DM, CAD	Headache, TMD
Tongue test	Normal sensation	Normal sensation	Normal sensation	Normal sensation	Normal sensation	Normal sensation
Facial test	Preauricular hypoesthesia	Preauricular hypoesthesia	Paresthesia	Preauricular hypoesthesia	Preauricular hypoesthesia	Preauricular hypoesthesia
Taste test	Normal, salt taste trigger	Normal, other than triggers	Normal	Normal	Hyperesthetic on same side	Hypoesthesia, triggers
Saliva	Clinically normal	Clinically normal	Clinically normal	Clinically normal	Clinically normal	Clinically normal
Psychologic factors	No	No	No	No	No	No
Ineffective medications	Neurontin, clonazepam	No treatment received	Tegretol, neurontin, klonopin	TCAs, anticonvulsants	Tegretol	Tegretol, neurontin, TCAs
Effective medications	None	None	None	None	None	Klonopin (2–3 weeks)

TMD = temporomandibular disorder; HTN = hypertension; DM = diabetes mellitus; CAD = coronary artery disease; TCA = tricyclic antidepressant.

Positive symptoms
- Onset of new pain follows surgery in region of ATN
- Pain occurs when triggered and consists of brief, paroxysmal episodes
- Pain is electric, lancinating in quality, and severe in intensity
- Pain centered in cutaneous sensory distribution of ATN
- Pain triggered by taste stimuli applied to ipsilateral anterior tongue

Negative symptoms
- Minimal or no pain between lancinating attacks
- Jaw movement does not provoke the lancinating pain
- Refractory period after pain occurs while subject continues to eat
- Psychologic factors not prominent features of presentation

Fig 1 Postive and negative symptoms of gustatory neuralgia.

Positive signs
- Preauricular sensory loss
- Preserved touch sensation on tongue

Negative signs
- No active anatomic pathology that would explain symptoms
- Touch sensation on tongue does not provoke pain
- No clinical dysautonomia (no facial flushing or sweating)

Fig 2 Positive and negative signs of gustatory neuralgia.

Similarities to TN
- Lancinating, electric paroxysms of pain
- No major pain between attacks
- Non-noxious sensory trigger
- Unilateral trigeminal distribution
- Autonomic and myofascial features are minor

Differences from TN
- Location of pain in ATN sensory region
- Preauricular hypoesthesia
- Prior trauma to ATN region common
- Gustatory trigger on ipsilateral tongue
- Little, if any, response to anticonvulsant/neuropathic drugs

Fig 3 Comparison of gustatory neuralgia to trigeminal neuralgia.

Discussion

The present report documents a series of patients who developed an unusual form of facial pain following surgery in the region of the ATN. The pattern of symptoms and clinical findings is consistent across the cases reviewed, and we propose that they constitute a unique syndrome. The syndrome is characterized by positive and negative symptoms and signs (Figs 1 and 2). This syndrome is distinct from other forms of clinical facial pain. We propose the name gustatory neuralgia (GN) for this disorder, to emphasize the neurogenic quality of the pain and the characteristic taste trigger.

Although the location and lancinating quality of pain are similar to those described for trigeminal neuralgia (TN), there are important differences between GN and TN that justify a separate designation (Fig 3). Trigeminal neuralgia is in most cases an idiopathic disorder that rarely results from trauma. The pain is electric and lancinating, usually radiating from the second or third trigeminal division. By definition there is no area of sensory loss. In contrast, the pattern of pain in GN is in the distribution of the ATN, and there was a temporary or permanent loss of sensation in this distribution in all of the present cases. A trigger zone in the perioral region is often seen in TN. Within this distribution light touch can evoke immediate, lancinating attacks of pain. In TN the trigger zone may be present in the mouth either on the lip or along the gum line; a consistent trigger zone on the tongue exclusively is extremely uncommon. In contrast, the trigger zone in GN is persistently present on the tongue, and taste is the only effective sensory stimulus. Finally, TN is very sensitive to anticonvulsants in most cases, and many authors consider a clinical response to carbamazepine to be diagnostic. None of the patients in the present series responded to anticonvulsants. These observations strongly support the view that GN is a distinct entity with only a superficial similarity to TN.

Trauma to the ATN during surgery is the probable cause of GN. This hypothesis is consistent with the postsurgical nature of the disorder, and with the distribution of the pain and the presence of ATN sensory loss. Parasympathetic efferent fibers for salivation travel within the ATN, along with cutaneous sensory afferents. The clinical findings in GN may result from an abnormal interaction between salivatory efferent fibers and trigeminal sensory afferents within the traumatized nerve,

with taste stimuli activating salivatory efferents. Because of this pathologic rewiring, taste stimuli could trigger paroxysmal pain in the distribution of ATN sensory fibers in GN patients.

A number of other hypothetical peripheral or central neurogenic mechanisms could explain the observed clinical findings in GN. Truax[22] hypothesized that this syndrome is caused by interruption of the sympathetic vasoconstrictor fibers to the parotid gland, resulting in unopposed, parasympathetic mediated vasodilation in response to gustatory stimuli. This theory seems unlikely, since it predicts that pain would result from a variety of forms of sympathetic blockade; this is not the case.[25] Sharav et al[23] proposed that the sensory characteristics in their case were "very similar to those of idiopathic trigeminal neuralgia" and that the atypical characteristics could represent those seen in "pre-trigeminal neuralgia." They proposed that many of the characteristics present in their case "clearly indicated central mechanisms" that are similar to those proposed for TN.

De Benedittis[24] reviewed the current theories proposed for the ATN syndrome (Frey syndrome). In his discussion he hypothesized in several lines of indirect evidence that the pain in this syndrome might be a result of "partial deafferentation." Although central mechanisms may be important for both disorders, it is worth reemphasizing that there are distinct differences between GN and TN, and there remains no strong evidence for a predominant role of either a peripheral or central mechanism in either disorder.

Neuropathic pain in the trigeminal system is a significant medical problem. A variety of neuropathic pain syndromes have been described, and the symptoms are often persistent and severe, resulting in substantial pain-related impairment and disability for the affected individuals. Although these conditions occur frequently, there is little understanding of the underlying mechanisms, and treatments for these conditions (with the exception of TN) are often ineffective. In the present study, the authors described a series of patients with highly concordant symptoms—most consistent with trauma to the ATN—that involve both sensory and autonomic functions. Common features among the patients reported, the persistent nature of the symptoms, the reliable presence of a discrete trigger zone responsive to taste, and a defined region of abnormal sensation all provide potential avenues of investigation to delineate the nature of this neuropathic disorder. It should also be emphasized that much is known about the transmitter systems and the neural control of the parotid gland; since damage to the autonomic fibers that innervate this gland is a probable component of the syndrome, a rational approach to pharmacologic analysis of the disorder may be attainable. Such analysis would have clear therapeutic implications, not only for those patients who suffer from GN, but also for patients with neuropathic pain disorders in general.

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Resumen

Modelo Clínico del Dolor Neuropático Trigeminal: Neuralgia Gustatoria Post-Traumática

En este articulo se habla de seis casos en los cuales la queja principal fue el dolor facial recurrente, episódico que fue provocado por el estímulo del gusto. El dolor ocurrió después de unos días o semanas luego de la cirugía de cabeza y cuello. Los pacientes reportaron que el estímulo de la comida en la boca avocó el dolor episódico, parecido a un choque eléctrico en una localización preauricular del lado de la cirugía. El olor a comida o, aunque menos confiable, la excitación emocional también pueden estimular el dolor. Los movimientos mandibulares no avocaron el dolor, y entre los ataque lancinantes no había dolor o sólo una molestía ligera. Luego de un episodio de dolor, hubo un período refractario durante el cual el dolor no puedo ser producido. El examen físico demostró que existía una perdida censorial preauricular de distribución variable. No se observaron clínicamente señales vasomotoras o de transpiración anormal. No se encontró patología odontogénica, muscular, neurológica, psicológica, o de las glándulas salivares, que pudiera explicar los síntomas clínicos. No se alivió el dolor con las dosis estándar de anticonvulsivantes que son usados comúnmente pare tratar la neuralgia del trigémino. La duración de los síntomas del dolor recurrente en este grupo fue de 8 a 132 meses sin remisión. La neuralgia gustatoria puede ser un síndrome discreto que puede resultar de las interacciones anormales entre las fibras eferentes salivares y las fibras aferentes sensoriales trigeminales en el nervio auriculotemporal lesionado. Los rasgos únicos en su género de este desorden lo convierten en un modelo clínico potencialmente útil pare la investigación de las interacciones autonómicas/sensoriales en el dolor neuropático.

Zusammenfassung

Posttraumatische Geschmacksneuralgie: Ein klinisches Modell des Trigeminalen Neuropathischen Schmerzes

Sechs Fälle wurden berichtet, in welchen die ursprüngliche Beschwerde ein episodischer, wiederkehrender Gesichtsschmerz war, der wurde durch einen Geschmacksstimulus ausgelöst. Der Schmerz kam Tage bis Wochen nach Kopf- und Nackenchirurgie vor. Die Patienten berichteten, dass ein im Mund plazierter Nahrungsstimulus episodische, elektroschock-ähnliche Schmerzen an einer präaurikulären Stelle auf der chirurgischen Seite hervorrief. Der Geruch von Nahrung oder, weniger zuverlässig, emotionale Erregung konnten den Schmerz auch auslösen. Mandibuläre Bewegung rief den Schmerz nicht hervor, und zwischen den lanzinierenden Attacken war entweden kein Schmerz oder nur milder Diskomfort. Im Anschluss an eine Schmerzepisode gab es eine Refraktärperiode, während welcher der Schmerz nicht ausgelöst werden konnte. Die physikalische Untersuchung zeigte einen präaurikulären Sensibilitätsverlust von variabler Verteilung. Kein abnormales Schwitzen oder vasomotorische Befunde waren klinisch augenscheinlich. Keine odontogene, muskuläre, Speicheldrüsen-, neurologische oder psychologische Pathologie werde gefunden, um die klinischen Symptome zu erklären. Der Schmerz wurde nicht gelindert mit Standarddosen von Antikonvusiva, welche gewöhnlich verwendet werden, um Trigeminusneuralgien zu behandeln. Die Dauer der wiederkehrenden Schmerzsymptome in dieser Gruppe betrug 8 bis 132 Monate ohne Nachlassen. Die Geschmacksneuralgie könnte ein diskretes Syndrom sein, das aus abnormalen Interaktionen zwischen salivatorischen efferenten Fasern und trigeminalen sensorischen afferenten Fasern im beschädigten Nervus auriculotemporalis resultiert Die einzigartigen Merkmale der Erkrankung machen es zu einem möglicherweise nützlichen Modell zur Untersuchung von autonomen/sensorischen Interaktionen beim neuropathischen Schmerz.

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