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Pre-trigeminal neuralgia

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Article abstract—Eighteen patients who subsequently developed typical trigeminal neuralgia experienced a prodromal pain termed “pre-trigeminal neuralgia.” These patients described their prodromal pain as a toothache or sinusitis-like pain lasting up to several hours, sometimes triggered by jaw movements or by drinking hot or cold liquids. Typical trigeminal neuralgia developed a few days to 12 years later, and in all cases affected the same division of the trigeminal nerve. Six additional patients experiencing what appeared to be pre-trigeminal neuralgia became pain-free when taking carbamazepine or baclofen. Recognition of pre-trigeminal neuralgia makes it possible to relieve the pain with appropriate medications and avoid unnecessary irreversible dental procedures.

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Typical attacks of trigeminal neuralgia consist of brief paroxysms of excruciating pain limited to some part of the distribution of the trigeminal nerve and often triggered by stimulating an area of skin or mucous membrane.¹⁻³ However, not every patient starts out in this fashion. As first noted by Symonds,⁴ some patients experience a dull, continuous, aching pain in the upper or lower jaw at the onset of their illness, and only later develop the classic paroxysmal pain. Mitchell⁵ termed this prodromal pain “pre-trigeminal neuralgia.” Similar prodromal sensations have also been reported in some cases of glossopharyngeal neuralgia.⁶

Pre-trigeminal neuralgia presents a considerable diagnostic challenge, as it is easily confused with pain of dental origin. This report describes the clinical features of 24 patients who had pre-trigeminal neuralgia, with the aim of further delineating this disorder, its differential diagnosis, and its treatment.

Methods. Review of our records disclosed 18 patients suffering from typical trigeminal neuralgia who had a history of a different type of pain prior to the onset of their typical painful paroxysms (table 1). The diagnosis of trigeminal neuralgia was made according to the usual criteria.¹⁻³ Neurologic examination and CT or MRI of the head were normal.

Another 6 patients were seen while they were experiencing what appeared to be pre-trigeminal neuralgia (table 2). The diagnosis of pre-trigeminal neuralgia in these patients was based on the following criteria: (1) description of pain similar to the prodromal pain reported by the patients listed in table 1; (2) normal neurologic and dental examinations; (3) normal CT or MRI of the head; and (4) patients became pain-free with carbamazepine or baclofen.

Results. The 18 patients who were seen after they had developed typical trigeminal neuralgia (table 1) de-

scribed their prodromal pain as a toothache or sinusitis-like pain. These pains were triggered by chewing, drinking hot or cold liquids, brushing the teeth, yawning, or talking in 8 of the 18 patients. Typical trigeminal neuralgia developed a few months to 12 years later and involved the same branch of the trigeminal nerve in all cases. In 12 patients the prodromal pain evolved directly into typical trigeminal neuralgia attacks, while 6 patients experienced isolated episodes of prodromal pain with a pain-free period of 1 to 11 months before the onset of their typical paroxysms of trigeminal neuralgia.

Our patients' ages at the onset of their prodromal pain ranged from 22 to 81 years with a mean of 56.2 years. This is comparable with the age range for pre-trigeminal neuralgia and typical trigeminal neuralgia reported by Mitchell,⁵ as well as the large series of patients with trigeminal neuralgia reported by Harris⁷ and by Rushton and Olafson.⁸ There were 11 women and 7 men in our series, and the pain occurred almost evenly on the right or the left side (10:8).

The patients in table 2 did not have typical trigeminal neuralgia but described a similar pain to the prodromal pain reported by the patients listed in table 1. This pain was relieved by carbamazepine or baclofen and had no other discernible cause. The age range of these patients at the onset of their pain was 37 to 71 years with a mean of 52.9 years. There were 4 women and 2 men in this group, and the pain occurred evenly on the right or the left side.

Case reports. Patient 1. A 68-year-old retired teacher had suffered from episodes of toothache-like pain in her left lower jaw for 6 months. These episodes would last several minutes and were precipitated by brushing her teeth, eating, or talking. The frequency and severity of these episodes gradually in-

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Table 1. Patients with typical trigeminal neuralgia who had a medical history of pre-trigeminal neuralgia (PTN)

Pt no.	Age (yrs)	Sex	Duration of PTN	Description of PTN pain	Location of PTN
1	68	F	6 mos	Toothache	L-V3
2	76	M	14 mos	Dull	L-V3
3	30	F	16 mos	Tingling	L-V1, 2
4	72	M	6 mos	Dull throbbing toothache	R-V3
5	82	M	3 mos	Aching	R-V1, 2
6	67	F	2 yrs	Dull, sharp	R-V2, 3
7	67	M	2 yrs	Dull aching	R-V3
8	58	M	10 yrs	Dull	L-V3
9	53	M	4 yrs	Deep aching	L-V3
10	34	F	12 yrs	Toothache + some sharp pains	L-V2
11	57	M	5 yrs	Aching	R-V3
12	52	F	7 mos	Toothache	R-V3
13	55	F	3 mos	Constant upper jaw toothache	R-V2
14	65	F	3 yrs	Dull aching	R-V1, 2
15	67	F	2 yrs	Gnawing	L-V3
16	42	F	4 yrs	Aching	L-V3
17	43	F	9 mos	Burning	R-V1
18	25	F	3 mos	Dull toothache	R-V2

creased and prompted extensive dental evaluations. Her regular dentist could find no evidence of a dental pathologic condition and referred her to an endodontist. The endodontist noted that the patient complained of a "bright, pricking, steady pain" which was triggered by touching her lower lip and by jaw or tongue movements. As there was no evidence of endodontic disorder, she was referred to an oral surgeon. The oral surgeon discovered a mass in her left neck which was found to be a vagal body tumor and which was excised uneventfully on April 1, 1987.

Postoperatively, the patient was pain-free for about 1 month, but on May 14 she had a sudden onset of flashes of severe stabbing pain in her left lower jaw, precipitated by talking, eating, or brushing her teeth. By the time she was admitted to Presbyterian-University Hospital, she had lost 14 lbs because any attempt to eat was likely to trigger an excruciating attack of pain.

Neurologic examination showed a somewhat elderly woman in acute distress who was very reluctant to talk and who was experiencing repeated painful paroxysms. No other abnormality was noted. CT and CSF examination were both normal.

Baclofen decreased the attacks of pain. However, the next day she became acutely confused, which cleared after baclofen was discontinued. She was then given carbamazepine and became pain-free taking 100 mg tid. She is now doing well with only 100 mg bid. Attempts to discontinue carbamazepine have resulted in a recurrence of the attacks of pain.

Patient 10. In 1968-1969 a 34-year-old woman developed an aching pain in her left cheek lasting minutes to hours when she would chew or yawn. In 1976, she had her teeth ground down in an attempt to relieve what was thought to be temporomandibular joint dysfunction, but this procedure did not help. She then underwent 3 years of psychotherapy which did not help. In 1982, she was treated with an intraoral orthosis and was pain-free for 7 to 8 months, but then the pain recurred. In September 1985, she had a left temporomandibular joint ar-

Table 2. Patients seen while they had pre-trigeminal neuralgia (PTN)

Pt no.	Age (yrs)	Sex	Duration of PTN	Description of PTN pain	Location of PTN
19	38	M	1 yr	Dull toothache	L-V3
20	63	F	20 yrs	Sharp	R-V2, 3
21	43	F	6 yrs	Sharp, burning	R-V2, 3
22	60	M	5 yrs	Burning	R-V2
23	69	F	1 yr	Aching	L-V2
24	81	F	10 yrs	Steady aching	L-V2

Pain relieved by carbamazepine in patients 19-22 and by baclofen in patients 23 and 24.

throplasty with meniscorrhaphy and meniscoplication. Postoperatively, she experienced left facial paralysis and left facial numbness. The facial palsy gradually resolved over the next few months but she continued to experience some numbness over her left cheek.

In August 1986, she began to suffer from electric shock-like sensations "like lightning," in her left eye and cheek, triggered by finching or suddenly moving her head. She also experienced burning and stinging in the left maxillary and frontal regions. She was given carbamazepine in December 1986, which stopped the flashes of pain at a dose of 1,000 mg/d but caused sleepiness, constipation, and menstrual irregularity. It was then found that baclofen 30 mg/d also controlled the flashes of pain, but caused a persistent cough for which no other cause could be found and which stopped within 48 hours after baclofen was discontinued. She then resumed taking a lower dose of carbamazepine (600 mg/d). On this regimen she experienced occasional flashes of pain and continued to complain of a dull aching pain "like she had been hit with a bat."

CT in April 1987 and MRI in January 1988 were normal. Neurologic examination was within normal limits except for a subjective diminution in pinprick and light touch perception over the left maxillary region.

Patient 20. A 63-year-old woman had a 20-year history of episodes of pain in her right face. She described these attacks as a sharp pain which came on gradually, starting in the upper and lower gums and then spreading to involve her whole right face, eye, and ear, and lasting up to 12 hours. The attacks were precipitated or exacerbated by alcohol.

Neurologic examination was normal. Dental examination showed her to be partially edentulous with marginal gingivitis around the remaining teeth, but no gross pathologic condition.

A trial of carbamazepine 100 mg tid 12 years after the onset of her pain produced complete relief. Carbamazepine was discontinued after 1 year, and she remained pain-free without medication for 2 years. When the pain recurred, it was again relieved by carbamazepine, but she required 200 mg tid with a serum level of 7.6 µg/ml. Two years later she experienced another recurrence, which was only partially relieved by carbamazepine 300 mg tid. The addition of phenytoin at that time made her pain-free except for occasional twinges when chewing. This regimen resulted in random serum levels of 7.8 µg/ml carbamazepine (therapeutic range, 4.0 to 12.0 µg/ml) and 5.5 µg/ml phenytoin (therapeutic range, 10.0 to 20.0 µg/ml).

The spontaneous onset of the attacks with no local triggering stimulus, the extensive spread of the pain to include even the aural area, the many hour duration of the attacks, and the provocation by alcohol are strongly suggestive of migraine or

cluster headaches, but the consistent relief with carbamazepine and phenytoin warrants, we think, the diagnosis of pre-trigeminal neuralgia as well. An association between pre-trigeminal neuralgia and cluster headaches is not surprising since there have been a number of reports of an association between trigeminal neuralgia and cluster headaches.⁹

Discussion. In their classic papers on trigeminal neuralgia, Fothergill^{10,11} and Pujol¹² noted the importance of distinguishing this disorder from pain secondary to dental disease to avoid "the useless and unnecessary extraction of entire rows of healthy teeth."¹² This differential diagnosis is sometimes difficult in the atypical patient who reports long periods of constant aching with superimposed stabs of sharper pain¹³ and can be exceedingly difficult in those patients whose trigeminal neuralgia starts out in an atypical fashion.^{4,5}

The proper evaluation of patients complaining of chronic, not readily explained, orofacial pain requires a careful history, a detailed neurologic and dental examination, and CT or MRI of the posterior and middle fossa. The greater anatomic detail and morphologic specificity provided by MRI¹⁴⁻¹⁷ appear to make it the preferable imaging procedure to rule out posterior fossa tumors that may cause facial pain.^{18,19} Pre-trigeminal neuralgia must be differentiated from trigeminal pain due to small tumors, atypical facial pain or atypical odontalgia, facial or "lower half" migraine, toothache of pulpal origin, sinusitis, and myofascial pain or temporomandibular dysfunction.

Atypical odontalgia is a poorly understood phenomenon which may be a variant of atypical facial pain.²⁰⁻²² Patients complain of chronic aching, burning, or throbbing pain in apparently normal teeth and surrounding alveolar bone. The pain is not relieved by local anesthetic blocks or antiepileptic drugs. Like atypical facial pain, it is often associated with depression and may respond to treatment with tricyclic antidepressants.

Temporomandibular joint dysfunction and myofascial pain is an overdiagnosed, but nevertheless real, entity which also causes chronic aching orofacial pain.^{23,24} These patients complain of pain on moving their jaws or palpation of the masticatory muscles, and show limitation of jaw opening, deviation of the mandible on opening or closing, and noises over the temporomandibular joint.

Recognizing that some patients may experience a dull, continuous, aching pain in the upper or lower jaw (pre-trigeminal neuralgia) before developing the characteristic paroxysmal attacks of typical trigeminal neuralgia makes it possible to relieve the pain with appropriate medication and to avoid unnecessary, and often destructive, dental procedures. Our observations also suggest that reports of trigeminal neuralgia occurring after dental procedures, such as tooth extractions or endodontic procedures, may indeed represent instances of pre-trigeminal neuralgia that was misdiagnosed as dental disease.

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References

1. White JC, Sweet WH. Pain and the neurosurgeon. Springfield, IL: Charles C Thomas, 1969:123-178.
2. Lance JW. Mechanism and management of headache, 4th ed. London: Butterworths, 1982:58-60.
3. Selby G. Diseases of the fifth cranial nerve. In: Dyck PJ, Thomas PK, Lambert EH, Bunge R, eds. Peripheral neuropathy, 2nd ed. Philadelphia: WB Saunders, 1984:1224-1265.
4. Symonds C. Facial pain. *Ann R Coll Surg Engl* 1949;4:206-212.
5. Mitchell PG. Pre-trigeminal neuralgia. *Br Dent J* 1980;149:167-170.
6. Rushton JG, Stevens C, Miller RH. Glossopharyngeal (vagus-glossopharyngeal) neuralgia: a study of 217 cases. *Arch Neurol* 1981;38:201-205.
7. Harris W. An analysis of 1,433 cases of paroxysmal trigeminal neuralgia (trigeminal tic) and the end-results of gasserian alcohol injection. *Brain* 1940;63:209-224.
8. Rushton JG, Olafson RA. Trigeminal neuralgia associated with multiple sclerosis: report of 35 cases. *Arch Neurol* 1965;13:383-386.
9. Lance JW. Mechanism and management of headache, 4th ed. London: Butterworths, 1982:205-226.
10. Fothergill J. Observations on the use of hemlock. Medical observations and inquiries by a society of physicians, London. 1769;3:400-418.
11. Fothergill J. Of a painful affection of the face. Medical observations and inquiries by a society of physicians, London. 1773;5:129-142.
12. Pujol M. Essai sur la maladie de la face nommée le tic douloureux. Paris: Theophile Barrois, 1787.
13. Sweet WH. The treatment of trigeminal neuralgia (tic douloureux). *N Engl J Med* 1986;315:174-177.
14. Lye RH, Ramsden RT, Stack JP, Gillespie JE. Trigeminal nerve tumor: comparison of CT and MRI—case report. *J Neurosurg* 1987;67:124-127.
15. Rigamonti D, Spetzler HE, Shetter A, Drayer BP. Magnetic resonance imaging and trigeminal schwannoma. *Surg Neurol* 1987;28:67-70.
16. Tanaka A, Takaki T, Maruta Y. Neurinoma of the trigeminal root presenting as atypical trigeminal neuralgia: diagnostic values of orbicularis oculi reflex and magnetic resonance imaging. *Neurosurgery* 1987;21:733-736.
17. Yuh WTC, Wright DC, Barloon TJ, Schultz DH, Sato Y, Cervantes CA. MR imaging of primary tumors of trigeminal nerve and Meckel's cave. *AJNR* 1988;9:665-670.
18. Bullitt E, Tew JM, Boyd J. Intracranial tumors in patients with facial pain. *J Neurosurg* 1986;64:865-871.
19. Nguyen M, Maciewicz R, Bouckoms A, Poletti C, Ojemann R. Facial pain symptoms in patients with cerebellopontine angle tumors: a report of 44 cases of cerebellopontine angle meningioma and a review of the literature. *Clin J Pain* 1986;2:3-9.
20. Reik L Jr. Atypical odontalgia: a localized form of atypical facial pain. *Headache* 1984;24:222-224.
21. Graff-Radford SB, Solberg WK. Atypical odontalgia. *Calif Dental Assoc J* 1986;14:27-32.
22. Solberg WK, Graff-Radford SB. Orofacial considerations in facial pain. *Semin Neurol* 1988;8:318-323.
23. Greene CS. Temporomandibular joint disorders, chap 37. In: Clark JW, ed. Clinical dentistry, vol 2. Philadelphia: Harper & Row, 1984.
24. Heft MW. Prevalence of TMJ signs and symptoms in the elderly. *Gerontology* 1984;3:125-130.

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