

Unnecessary Extractions in Patients with Hemicrania Continua: Case Reports and Implication for Dentistry

Sanjay Prakash, DM

Associate Professor
Department of Neurology
Medical College
Baroda, Gujarat, India

Nilima D. Shah, MD

Senior Resident
Department of Psychiatry
Medical College
Baroda, Gujarat, India

Bhavna V. Chavda, MD

Resident
Department of Medicine
Medical College
Baroda, Gujarat, India

Correspondence to:

Dr Sanjay Prakash
O-19, doctor's quarter, jail road
Baroda, Gujarat, India, 390001
Fax no: (91) (2692) 258700
Email: drprakashs@yahoo.co.in

Headache and facial pain are both very high in the general population. Headache has been identified as one of the associated conditions in patients with chronic orofacial pain. The interrelation between the two has not been explored in the literature. Patients with facial pain often initially seek the care of a dentist. Misdiagnosis and multiple failed treatments (including invasive procedures) are very common in this population. This case report describes four patients whose condition fulfilled the International Headache Society's criteria for hemicrania continua but whose teeth were extracted because their pain was suspected to be of odontogenic origin. Each patient's records and the literature were reviewed for possible reasons for the unnecessary extractions. The findings suggest that initial treatment with drugs specific for primary headache disorders should be instituted before subjecting patients to invasive procedures. J OROFAC PAIN 2010;24:408-411

Key words: facial pain, headache, hemicrania continua, indomethacin, indomethacin responsive headaches

Both chronic daily headache and chronic orofacial pain are very high in the general population. About 5% of the adult population suffer from headache on a daily or near-daily basis.¹ Facial pain affects at least 10% of the adult and 50% of the elderly population.² The defining boundary between facial pain and headache is the orbitomeatal line, but there is no anatomical or physiological basis for this distinction. Facial pain and headache disorders frequently cross this boundary and can lead to a diagnostic dilemma. The presence of headache as a comorbid condition in patients with facial pain or vice-versa is another factor for creating diagnostic confusion.³ Besides these, there are many primary headache disorders that may exclusively present as orofacial pain (including isolated tooth pain).⁴⁻¹¹ Patients with orofacial pain often seek the care of dental practitioners and oral surgeons. These patients are frequently subjected to various invasive dental and other procedures.¹² The high prevalence of dental caries and periodontal diseases in the population may further complicate the diagnostic and therapeutic decisions.¹³ This article reports four patients with hemicrania continua (collected over 2 years) in whom teeth were unnecessarily extracted. Written informed consent was obtained from all the patients in order to publish the report.

Case Reports

Case 1

A 61-year-old male presented with a 22-year history of continuous left-sided headache and facial pain. The pain was described as a steady ache of mild to moderate severity with superimposed exacerbations of incapacitating and pulsatile pain. The pain was maximal in the supraorbital and infraorbital areas, with radiation to the whole of the face, including teeth, on the same side. The usual duration of the exacerbations was 2 to 4 hours. The patient experienced one to two exacerbations per week. Nocturnal attacks were common. Cranial autonomic symptoms (feeling of sand in the eye, conjunctival injection, and lacrimation) were noted in about one tenth of the exacerbations. However, the presence of autonomic features was never investigated by any treating physician in the past. The exacerbations were not associated with nausea, vomiting, photophobia, phonophobia, etc.

The patient visited several dentists, general physicians, neurologists, and otolaryngologists. A number of different diagnoses were made (atypical facial pain, atypical odontalgia, sinusitis, etc). He underwent sinus surgery, four root canal treatments, and six extractions (teeth number 25–28, 36–37). However, these procedures never provided symptomatic relief. He received numerous pain killers, antibiotics, and antihistamines. A few pain killers (details not available) provided some relief to his symptoms. However, relief was never complete, and the pain persisted.

A diagnosis was made of possible hemicrania continua. Indomethacin was started, and the patient showed complete relief of symptoms to 75 mg three times a day for 5 days. Skipping the drug resulted in recurrence of pain, and there was no attempt to lower the dose of indomethacin in the next 10 months of follow-up.

Case 2

A 45-year-old male presented with a 3-year history of continuous pain on the left side of the whole face and forehead. The pain was described as a continuous dull ache of mild to moderate severity with superimposed exacerbations of throbbing pain. The pain of exacerbation was maximal in the left supraorbital and infraorbital areas. The exacerbations occurred one to two times per week, lasting from 30 minutes to 2 hours. The pain never occurred on the right side. There were occasional nocturnal exacerbations. The exacerbations were associated with conjunctival injection and/or rhinorrhea in about 50% of the attacks. However, this part of the history was never investigated or reported on previous consultations. The exacerbations were not

associated with nausea, photophobia, or phonophobia. The patient also had a history of toothache on the left side. The pain in the teeth was intermittent (6 to 24 hours) and was not related to the exacerbations of the headache and facial pain. His previous diagnoses were caries, pulpitis (teeth number 26–28), atypical odontalgia, atypical facial pain, sinusitis, psychiatric disorders, etc. Magnetic resonance imaging (MRI) of the brain and sinuses was performed on a few occasions in the past, and scans were normal. The patient underwent root canal therapies and extractions (teeth number 26–28). The toothache was relieved on a few occasions by the dental procedures, but it did not have any effect on the facial pain.

The authors made a diagnosis of possible hemicrania continua. Indomethacin (50 mg, three times a day) was prescribed and resulted in complete response. Attempts to reduce the dose in the follow-up period (up to about 12 months) were not successful.

Case 3

A 48-year-old woman had a history of toothache about 2 years ago. Her right upper wisdom tooth (number 18) was extracted for caries. Her pain subsided completely within a few days. However, about 2 months later, she felt a continuous dull ache of mild to moderate severity in the right maxillary area. She had exacerbations of more severe pain, occurring three to four times a month, and each lasting for 4 to 12 hours. The exacerbations were maximal in the right maxillary area with radiation to whole right side of the face and frontotemporal areas. Occasionally, the exacerbations were associated with dental pain in the right maxillary area. The patient noted nausea and/or photophobia during a few of the exacerbations. She had a number of diagnoses: atypical odontalgia, atypical facial pain, sinusitis, chronic migraine, atypical facial pain with migraine, etc. Trials with various medications produced minimal or no effect. Even trials with sumatriptan and rizatriptan during the exacerbations did not provide relief. Two other teeth (number 16–17) were extracted from the right maxilla on the assumption that these were the source of the pain as she had evidence of caries in these teeth. However, this procedure resulted in no relief. The patient was referred to the authors' outpatient clinic for a possible neurological cause. An additional history of "feeling of sand in the eye," conjunctival injection, and nasal blockage on the left side was obtained during the exacerbations. MRI of the brain was normal. A diagnosis was made of possible hemicrania continua, and indomethacin was started. The pain subsided completely by indomethacin (50 mg, three times a day). Weaning off indomethacin was done on two occasions over the 6 months of follow-up but was not successful.

Case 4

A 38-year-old female presented with a 15-month history of right-sided headache and facial pain. The pain was initially described as a constant ache of mild (occasionally moderate) intensity in the right maxillary area (occasionally in the teeth). The initial diagnosis was sinusitis. However, it was not affected by various analgesics and antibiotics. The dental examination at that time was suggestive of caries (teeth number 16–18). The patient was subjected to root canal therapy (tooth number 18). However, there was no improvement. Over time, the pain extended to involve the right side of the face and frontotemporal areas. Superimposed on the background pain, the patient felt exacerbations of more severe excruciating pulsating pain. The exacerbations occurred two to 25 times daily, lasting from a few seconds to 10 minutes. Nocturnal exacerbations were common. The patient denied any presence of nausea, vomiting, photophobia, phonophobia, or auras. There were no triggering factors. She received a number of diagnoses for her symptoms: trigeminal neuralgia, atypical odontalgia, atypical facial pain, etc. Trials with various medications produced minimal or no benefit. One more tooth (number 17) was extracted from the right maxilla on the assumption that this was the source of the pain. MRI of the brain was normal.

The patient was referred to the authors' outpatient clinic for a possible neurological cause. The patient denied the presence of any cranial autonomic features, but the authors noted conjunctival injection during a few of the exacerbations. On further inquiry, during an ongoing attack, the patient admitted to having a "feeling of sand in the eye" and nasal stuffiness. A diagnosis was made of possible hemicrania continua, and a trial with indomethacin at a dose of 75 mg three times a day resulted in complete relief of symptoms. Skipping of the drug resulted in recurrence of pain in the next 6 months of follow-up.

Discussion

Misdiagnosis is very common for both chronic orofacial pain and primary headache disorders. A misdiagnosis of hemicrania continua is probably highest among all primary headache disorders.¹⁴

All four patients fulfilled the diagnostic criteria of the International Headache Society (IHS) for hemicrania continua.¹⁵ The diagnostic criterion for hemicrania continua overlaps with the criteria for trigeminal autonomic cephalalgias. However, the site of pain and the duration and frequency of the attacks (exacerbations) are not described in the criteria for hemicrania

continua.¹⁵ Therefore, if a patient or doctor focuses on the painful exacerbations and fails to consider the continuous background pain, hemicrania continua may be mistaken for cluster headache, paroxysmal hemicrania, migraine, trigeminal neuralgia, etc. In a study by Peres et al, about 70% patients with hemicrania continua met the IHS criteria for migraine.¹⁶ Rossi et al reported that 32% of patients with hemicrania continua fulfilled the criteria for cluster headache during exacerbations.¹⁴ These observations highlight the need for careful history-taking regarding the presence of continuous background headache.

In the same way, if we focus on the continuous background headache and fail to consider painful exacerbations, hemicrania continua may be mistakenly for atypical facial pain, atypical odontalgia (especially if the pain is localized below the orbitomeatal line), or temporomandibular disorder. Atypical odontalgia is classically defined as presence of continuous pain in a tooth or tooth site. However, it may spread to involve the entire face, head, or neck. Atypical odontalgia is known to show fluctuation in intensity.¹⁷ Therefore, there is the possibility of making a wrong diagnosis of atypical odontalgia in a patient with hemicrania continua.

The frequency, intensity, and components of autonomic features are usually meager and of moderate dimension in patients with hemicrania continua (as compared to trigeminal autonomic cephalalgias).¹⁶ The patients themselves may not volunteer or remember about the presence of cranial autonomic features. Case 4 outlined above admitted to having cranial autonomic features only when asked specifically during an attack. Rossi et al reported a similar case in which an autonomic feature was noted after the initial visit.¹⁴ The presence of a large number of cases of hemicrania continua-like headache (responsive to indomethacin) without cranial autonomic features in the literature¹⁸ suggests that patients may not come forward with a history suggestive of autonomic features in their first visit. Therefore, a patient with refractory chronic orofacial pain should be asked to look for the features of cranial autonomic symptoms during exacerbations in follow-up for a possible diagnosis of hemicrania continua.

Unfamiliarity with hemicrania continua may be another reason for a misdiagnosis. Twenty neurologists and seven headache specialists failed to make a correct diagnosis in the study by Rossi et al.¹⁴ Cases 1, 2, and 3 above had a history of cranial autonomic features. However, these were never investigated previously by any treating physician.

The exact site of pain is not defined in the IHS criteria for hemicrania continua.¹⁵ Available data suggest that the anterior part of the head may be

Table 1 Literature Review of Hemicrania Continua Misdiagnosed as Temporomandibular Disorders (TMD) or Dental Disorders

Authors	No. of cases	Main pain site	Misdiagnosis	Unnecessary treatments
Rossi et al ¹⁴	5*	NA*	TMD/dental pain	Dental extraction = 5 TMD surgery = 2
Taub et al ⁸	2	Case 1: right mandibular area, molar, preauricular, ear. Case 2: left ear, jaw, neck, tempofrontoparietal areas.	TMD TMD	Not any specific Not any specific
Alonso et al ⁷	1	Right maxillary premolar, temple, and vertex.	Dental pain	Dental extraction
Benoliel et al ⁹	1	Right head, face, mouth, maxillary molars.	Dental pain	Dental treatment (not specified)

* = 5 out of 25 patients misdiagnosed as dental pain. The details of individual patients were not available.

involved more frequently.¹⁶ However, it may radiate to involve the whole of the head and face. It may radiate even to teeth, temporomandibular joint, ear, and throat. Toothache or temporomandibular joint pain may be a presenting complaint in patients with hemicrania continua.^{7-11,19} This may lead to misdiagnosis of hemicrania continua as a dental or temporomandibular disorder, and the patient may be subjected to various erroneous treatments (Table 1).

The present case reports and review of the literature suggest that orofacial pain with or without headache may be a feature of hemicrania continua, and it should be a differential diagnosis in patients with chronic orofacial pain. The identification of these patients is important, as they respond very well to specific therapy. Trials should be made with the drugs effective for various primary headache disorders before subjecting such patients for surgical interventions. This will prevent unnecessary extraction of teeth in a subset of patients with primary headache disorders whose predominant complaint is chronic orofacial pain.

References

- Castillo J, Muñoz P, Guitera V, Pascual J. Epidemiology of chronic daily headache in the general population. *Headache* 1999;39:190-196.
- Madland G, Feinmann C. Chronic facial pain: A multidisciplinary problem. *J Neurol Neurosurg Psychiatry* 2001;71:716-719.
- Dando WE, Branch MA, Maye JP. Headache disability in orofacial pain patients. *Headache* 2006;46:322-326.
- Balasubramanian R, Klasser GD, Declanho R. Trigeminal autonomic cephalalgias: A review and implication for dentistry. *J Am Dent Assoc* 2008;139:1616-1624.
- Obermann M, Muller D, Yoon M-S, Pageler L, Diener H, Katsarava Z. Migraine with isolated facial pain: A diagnostic challenge. *Cephalalgia* 2007; 27:1278-1282.
- Moncada E, Graff-Radford SV. Benign indomethacin responsive headache presenting in the orofacial region: Eight case reports. *J Orofac Pain* 1995;9:276-284.
- Alonso AA, Nixdorf DR. Case series of four different headache types presenting as tooth pain. *J Endod* 2006;32:1110-1113.
- Taub D, Stiles A, Tucke AG. Hemicrania continua presenting as temporomandibular joint pain. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2008;105:e35-e37.
- Benoliel R, Robinson S, Eliav E, Sharav Y. Hemicrania continua. *J Orofac Pain* 2002;16:317-325.
- Iordanidis T, Sjaastad O. Hemicrania continua: A case report. *Cephalalgia* 1989;9:301-303.
- Trucco M, Antonaci F, Sandrini G. Hemicrania continua: A case responsive to piroxicam-beta-cyclodextrin. *Headache* 1992;32:39-40.
- Israel HA, Ward JD, Horrell B, Scrvani SJ. Oral and maxillofacial surgery in patients with chronic orofacial pain. *J Oral Maxillofac Surg* 2003;61:662-667.
- Patro BK, Kumar BR, Goswami A, Prakash Mathur VP, Nongkynrih B. Prevalence of dental caries among adults and elderly in an urban resettlement colony of New Delhi. *Indian J Dent Res* 2008;19:95-98.
- Rossi P, Faroni J, Tassorelli C, Nappi G. Diagnostic delay and suboptimal management in a referral population with hemicrania continua. *Headache* 2009;49:227-234.
- Headache Classification Subcommittee of the International Headache Society. The international classification of headache disorders, ed 2. *Cephalalgia* 2004;24 (suppl 1):1-160.
- Peres MF, Silberstein SD, Nahimas S, et al. Hemicrania continua is not that rare. *Neurology* 2001;57:948-951.
- Melis M, Lobo SL, Caroline Ceneviz C, et al. Atypical odontalgia: A review of the literature. *Headache* 2003;43:1060-1074.
- Prakash S, Shah ND, Bhanvadia RJ. Hemicrania continua unresponsive or partially responsive to indomethacin: Does it exist? A diagnostic and therapeutic dilemma. *J Headache Pain* 2009;10:59-63.
- Newman LC, Lipton RB, Solomon S. Hemicrania continua: Ten new cases and a review of the literature. *Neurology* 1994;44:2111-2114.