Odontalgia in Vascular Orofacial Pain

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A case of episodic, spontaneous odontalgia, aggravated by ingestion of cold food, with no apparent dental pathology is presented. Attempts at alleviating the pain by means of root canal treatment had failed in previous, similar episodes, and pain and pulpal hyperalgesia had shifted to other locations. Primary vascular orofacial pain was diagnosed and effective control obtained by prophylactic treatment with propranolol, a beta-adrenergic blocker. A prophylactic attempt with nifedipine, a calcium channel blocker, failed to alleviate the pain. This diagnostic entity and possible therapeutic approaches are discussed.
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Odontalgia that is evoked by the ingestion of cold foods and beverages is usually associated with pulpal pathology, most commonly caries. When pain becomes severe and spontaneous, a pulpal inflammatory process is suspected.[1] In these latter cases the affected tooth can be identified by means of clinical and radiographic examinations. The tooth is hyperalgesic to cold application, often tender to percussion, and may demonstrate a deep carious lesion, primary or secondary; radiographic examination may reveal a deep restoration.[2] Tooth pulp extirpation and endodontic therapy are usually sufficient to terminate the pain.[2]

However, there are cases that present pain characteristics similar to those associated with a pulpal inflammatory process, but with no apparent dental etiology. Endodontic tooth pulp extirpation in these cases will not abolish the pain. These cases are very puzzling to the dental practitioner, particularly when they do not respond to endodontic therapy. Several conditions that may mimic odontalgia are shown in Table 1. The experienced clinician will be able to diagnose most of these entities, especially since most of them have a major source of pain *extraorally* that refers *intraorally*. Moreover, local dental signs (thermal sensitivity, tenderness to percussion) are usually not present.

Odontalgia that is not due to pulpal pathology and does not fit any of the diagnoses presented in Table 1 has been called *phantom tooth pain, atypical odontalgia* (AO), or *atypical facial pain.*[1] It has been proposed that these are the result of vascular, migraine-like, pathophysiologic mechanisms.[3–5] While vascular mechanisms of odontalgia could not be confirmed by an attempt to correlate them with a history of migraine, a possible primary diagnosis of independent vascular orofacial pain is a suggested mechanism.[6]

Presented here is a case of spontaneous odontalgia aggravated by cold food ingestion. The pain was diagnosed as primary vascular odontalgia and controlled by chronic treatment with a beta-adrenergic blocker. Possible underlying pain mechanisms are discussed.

Table 1 Conditions that Mimic Odontalgia

Musculoskeletal
Arthralgia
Myalgia
Neuropathic
Trigeminal neuralgia and pretrigeminal neuralgia
Post-herpetic neuralgia
Phantom (deafferentation) toothache
Vascular
Vascular orofacial pain
Cluster headache
Paroxysmal hemicrania
Inflammatory
Papillitis/food impaction
Acute otitis media
Acute maxillary sinusitis
Obstructive sialoadenitis
Herpes zoster infection
Neoplastic
Local/metastatic tumors
Central nervous system lesions
Referred pain not from head and neck
Cardiogenic

Case Report

A 32-year-old female complained of 2 types of chronic orofacial pain. One, present on the right side of the face over the angle of the mandible, was constant, dull, and intensified by chewing. The other, on the left side, was daily and episodic, lasted from a few minutes to several hours, and was aggravated by ingestion of cold foods and drinks. This latter pain was severe and could wake the patient from sleep.

A vicious cycle of similar episodic pain attacks, which led to unsuccessful dental treatment, had begun 1 year previously. Several teeth had undergone root canal therapy and were then extracted. Dental treatment had provided only temporary relief, if any, but the pain always reappeared soon after in a different location, including the other side of the face. Nonsteroidal anti-inflammatory drugs provided short-term relief. Neurologic examination, including computed tomographic scanning of the head and base of skull, revealed no pathology.

On examination, the right temporomandibular joint and muscles of mastication were tender to palpation and the mouth opening was normal (48 mm interincisal). Intraoral examination revealed a well-maintained dentition and good oral hygiene. Extensive dental treatment had been performed in the past, including root canal therapy and extractions, but no active pathologic process was detected. Further tests revealed that the first and second left mandibular molars were hypersensitive to cold application (ethyl chloride), with marked overshoot, and were not tender to percussion. Intraoral radiographs revealed sound amalgam restorations, with no proximity to the pulp horns and no periapical pathology.

Based on the presenting symptomatology and the patient's past history, diagnoses of musculoskeletal pain on the right and vascular orofacial pain on the left were made. Treatment was initiated with 10 mg amitriptyline at night, increasing gradually to 35 mg, aimed at relieving both pain types. This resulted in relief of the musculoskeletal pain, but the patient still complained of marked sensitivity to cold in the above-mentioned teeth. We then decided to replace amitriptyline therapy with a beta-adrenergic blocking agent. Amitriptyline was reduced to 10 mg per day and propranolol (80 mg per day) was initiated. Blood pressure measurement at that time revealed moderate hypertension (160/110). Thus, in collaboration with the patient's physician, who continued monitoring her blood pressure, the authors gradually increased slow-release propranolol hydrochloride to 240 mg per day. Concomitantly, the amitriptyline was stopped, but this resulted in the reappearance of the right-sided musculoskeletal pain, and amitriptyline (10 mg per day) had to be re-initiated. Total pain relief was obtained, and the left mandibular molars were no longer sensitive to cold.

Upon her physician's suggestion, the authors decided at this stage to replace propranolol with another antihypertensive drug, nifedipine, a

Table 2 Comparison of Signs and Symptoms

Parameter	Disorder	
	Vascular orofacial pain	Irreversible pulpitis
Patient's report		
History of migraine	Sometimes	No
Response to past endodontic treatment	None	Complete relief
Accompanying autonomic signs	Common	None
Teeth affected are		
Hypersensitive to cold	Yes	Yes
Tender to percussion	No	Yes (80% of cases)
Changing in location	Common	Uncommon
Carious (clinical/radiologic)	Uncommon	Common
Response to therapy		
Endodontics	No relief	Complete relief
Nonsteroidal antiinflammatory drugs	Relief	Occasional relief
Amitriptyline	Relief	No relief
Beta-blockers	Relief	No relief

calcium channel antagonist. Although the patient remained normotensive, the left-sided, spontaneous, severe orofacial pain and dental sensitivity to cold returned. The patient was then transferred back to propranolol, and pain relief was again achieved.

Discussion

The odontogenic pain reported here clearly represents a diagnostic problem. The presenting complaint of severe spontaneous odontalgia aggravated by a thermal stimulus resembles irreversible pulpitis (Table 2). Diagnostic tests revealed hyperalgesia to cold stimuli, with lingering after-pain. The characteristics of the pain—namely spontaneous, severe, and poorly localized with a pulsatile quality—resemble those of pulpitis, which was the most tempting diagnosis.[2] However, the history of unsuccessful endodontic treatment followed by extraction aroused suspicion. A clear pattern of severe pain leading to radical treatment with no relief was reported. Moreover, the pain migrated from tooth to tooth and from side to side. The clinical and radiologic examinations showed 2 molars with deep amalgam restorations but without any obvious pathology. Additionally, 2 teeth were simultaneously sensitive and although it is possible that they were both affected by irreversible pulpitis, this is rare. Patients who additionally report long-lasting dental pain should arouse suspicion, since irreversible pulpitis usually has a short history.

Odontalgia with Vascular-Type Symptoms: Terminology

The International Association for the Study of Pain[7] has defined AO as severe pulsatile pain in the teeth without local pathology, and this entity may be similar to vascular odontalgia. In AO the patient has a history of extensive dental work without pain relief, and although root canal therapy or extraction may provide transient relief, the pain recurs.[8] The symptoms may start in one quadrant and spread across the midline to the opposite side.[8] Symptoms are reported to last from 1 month to 20 years, with an average of 3.1 years[8] (dental pain of such long duration should make clinicians wary). These symptoms were seen in our patient. The etiology of AO is unclear; however, vascular,[4] neuropathic,[9] or psychogenic[10] origins have been suggested. Atypical odontalgia may indeed demonstrate some characteristics of vascular-type pain in that it is pulsatile[11] and episodic.[8] Additionally, the pain does not always follow anatomic pathways of the peripheral nervous system,[12] and the effect of local anesthesia is variable, suggesting that the pain is not neuropathic in origin. At times, however, it is constant and has a burning quality,[9] indicating that neuropathic mechanisms may be involved, and this is best termed *neuropathic orofacial pain.* Thus it is possible that the clinical diagnosis of AO includes 2 different pathophysiologic entities, vascular and neuropathic.

Recently, a large group of patients with facial pain, including oral pain with vascular-type

symptoms, was described.[6] The patients suffered from episodic, severe pain that was usually unilateral and was accompanied by autonomic or systemic signs (55% of cases). A large number of these patients (38%) reported unsuccessful attempts at pain relief by dental treatment, but antimigraine-type therapies were reported to be effective. The term *vascular orofacial pain,* possibly a category of primary vascular-type craniofacial pain, was suggested. In a similar fashion, the history and symptomatology of our patient supported a diagnosis of vascular odontalgia, which was verified by the positive response to propranolol.

The pain quality and behavior of vascular toothache is similar to that of vascular-type headaches, ie, severe, unilateral, episodic pain that awakens the patient from sleep. Thus, a consideration of pain mechanisms in headache may be useful. Vascular-type headaches consist of migraine, cluster headache, and chronic paroxysmal hemicrania.[7] Migraine affects more women than men, and pain lasts for hours to days, accompanied by systemic signs such as nausea, photophobia, and phonophobia. Characteristic of some migraine headaches is the "aura"—forewarning signs that may be tactile, visual, or even olfactory and that precede the onset of headache. Autonomic signs are mainly associated with cluster headache and chronic paroxysmal hemicrania. In migraine, and sometimes in cluster headache, there is a tendency for pain to change sides; primary vascular odontalgia also demonstrates this behavior.

Moskowitz[13] has proposed that vascular-type headache is transmitted by intracranial perivascular sensory axons that originate largely from cell bodies within the trigeminal ganglia. Together with the intracranial vasculature, they constitute the trigeminovascular system.[13] When stimulated antidromically, these afferent nerve fibers promote a sterile inflammatory response by releasing vasoactive peptides at the nerve endings. The released peptides include substance P (SP), calcitonin gene-related peptide (CGRP), and neurokinin A, the most potent vasodilator being CGRP. The afferent fibers that release these neuropeptides are characteristic of C fibers. These neuropeptides cause vasodilatation of the intracranial vessels and increased permeability with plasma extravasation. Since neurogenic inflammation in the trigeminovascular system seems to play a central role in the genesis of vascular-type headaches, the same mechanism could function in the oral mucosa and teeth.

Possible Pathophysiology of Vascular Odontalgia

The oral tissues and the dental pulp receive their sensory innervation from the trigeminal nerve. The dental pulp receives fine unmyelinated nerve fibers that are C fibers and myelinated A-delta fibers.[14] In addition to sensory nerves, there are sympathetic fibers from the superior cervical ganglion. Nerve fibers exhibiting SP and CGRP-positive immunoreactivity have been demonstrated in the dental pulp and oral mucosa in several species.[15,16]

Theoretically, these could cause vasodilatation with increased vessel permeability in the oral tissues. Being rigidly encased within dentin, the pulp is a low-compliance environment, a situation similar to the skull encasing the brain. Thus, vasodilatation and increased vascular permeability will result in an increased pulpal hydrostatic pressure, which can cause dental hyperalgesia and spontaneous pain.

Treatment Strategy

Treatment strategy in vascular-type pain may be abortive and utilize ergot preparations or nonsteroidal anti-inflammatory drugs,[17] or alternatively, when pain is frequent, prevention of pain may be achieved by chronic intake of amitriptyline or adrenergic beta-blockers.[18]

In our patient, the symptoms and the dental history were those of vascular odontalgia. Pain frequency was high and favored prophylactic, rather than abortive, treatment. The patient responded favorably to treatment with beta-blockers, and this may give additional clues as to the underlying mechanism. Beta-blockers are presumed to be efficient in preventing migraine by blocking peripheral vasodilatory receptors, thus interfering with the vasodilatory phase in migraine. Beta-receptors are found in the pulpal vessels as well, which indicates that these may play a role in vascular permeability. Calcium channel antagonists are less efficient in migraine prophylaxis; this may explain why they were not successful in our patient. Amitriptyline has been shown to be effective in migraine[18] and in chronic craniofacial pain.[19] Although amitriptyline may be more effective than beta-blockers in migraine prophylaxis,[18] in our patient, amitriptyline was effective only in relieving the musculoskeletal pain. This may have been a dose-related effect.[6]

Conclusions

A case of a patient complaining of primary odontalgia with no detectable pulpal pathology is presented. The signs and symptoms mimicked pulpal pain and led to unwarranted treatment. Careful examination of patients with a thorough history usually will reveal the presence of vascular toothache that requires pharmacologic, and not operative, treatment.

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