

REVIEW ARTICLE



Atypical facial pain and atypical odontalgia: A concise review

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Abstract

The concept of pain has evolved from that of a one-dimensional sensation to that of a multi-dimensional experience encompassing sensory, discriminate, cognitive, motivational and affective qualities. Pain is the presenting symptom of a broad spectrum of diseases that needs to be diagnosed and treated. Establishing a precise diagnosis and providing effective treatment have become major challenges in medical and dental profession. Oro-facial pain is the field of dentistry devoted to the diagnosis and management of chronic, complex, facial pain and oro-motor disorders. Since different diseases produce characteristic patterns of tissue damage, the quality, time course, location of a patient's pain complaint and the location of tenderness provide important diagnostic clues and are used to evaluate the response to treatment. The general characteristics, etiologic characteristics, pathophysiology, differential diagnostic criteria, and therapeutic options of atypical facial pain and atypical odontalgia are described.

Keywords: Atypical, odontogenic, oro-facial, pain

Introduction

Pain gravely impairs the lives of millions of people around the world and is considered as a symptom of the disease to be diagnosed and treated. The most recent definition of pain produced by the task force on taxonomy of the International Association for the Study of Pain (IASP) is "An unpleasant sensory and emotional experience associated with actual or potential tissue damage or described in terms of such damage."^[1] Oro-facial pain encompasses a myriad of signs and symptoms within and outside the oral cavity. As a symptom it may be due to disease of structures, generalized musculoskeletal or rheumatic disease, peripheral or central nervous system disease or psychological abnormality or the pain may be referred from other sources.^[2] The possible causes of oro-facial pain are considerable and cross the boundaries of many medical and dental disciplines. An interdisciplinary approach is often required to establish a diagnosis and for treatment.

Establishing a precise diagnosis and providing effective treatment have become major challenges in medical and dental disciplines. The dentist has a great responsibility of proper management of pain in and around the face, oral cavity and neck. The dentist responsibility in managing the pain is two-fold, first being ability to diagnose the local and systematic causes and to identify those symptoms that are correctable by

dental therapy. The second responsibility of the dentist relates to therapy. Once the pain component is correctly identified as a condition amenable to dental therapy, treatment should be the next step.

Atypical facial pain (AFP) was first described by Frazier and Russell (1924),^[3] differentiating it from trigeminal neuralgia. It falls within the category of "Facial Pain Not Fulfilling Other Criteria" in the classification system of the International Headache Society.^[4] As this implies, the diagnosis is generally made after excluding local oro-facial disease, neurological disorders and related systemic diseases. It is not related with objective neurological, facial or oral findings and frequently presents with a non-anatomical and even a migratory pattern.^[5] Due to the vagueness of this term and in an attempt to avoid further confusion, the IASP discontinued to list AFP in their classification of chronic pain. Instead the broader term AFP has been replaced by two specific sub-entities namely "atypical odontalgia" (AO) and "glossodynia and sore mouth" (oral dysesthesia); the latter of these is also referred to as the "burning mouth syndrome."^[6]

Understanding of the condition is complicated further by a plethora of other terms, used synonymously or as variations of the theme. These include:

1. Phantom tooth pain (Marbach [1978];^[7] Marbach *et al.* [1982])^[8]

2. Atypical facial neuralgia (Marbach *et al.* [1982]^[8])
3. AO (Rees and Harris [1978]^[9])
4. Migratory odontalgia (Solomon and Lipton, [1990]^[10])
5. Wandering tooth syndrome (Kudrow *et al.* [1991]^[11]) and
6. Dental causalgia (Massler [1981]^[12]).

Because it is based on diagnosis by exclusion and is poorly understood this condition may be missed, resulting in excessive treatment, or over-diagnosed leading to inappropriate treatment. Thus, it lends itself to abuse.

Although similar to AFP, AO is better defined anatomically. AO, also known as idiopathic or phantom tooth pain, was first reported by McElin and Horton (1947).^[13] This clinical condition has been validated extensively, yet it is rarely reported. AO has been referred by Rees and Harris (1978)^[9] as tooth pain with no obvious organic cause. Lascelles (1966)^[14] regarded AO as a localized form of AFP. It is poorly understood phenomenon associated with persistent pain in apparently normal teeth and surrounding alveolar bone. Campbell *et al.* (1990)^[15] reported the epidemiologic information, which indicates that 3-6% of patients develop AO after endodontic treatment.

Etiology and Pathogenesis

The causes and pathophysiology of AFP and AO remain enigmas. To explain them, several casual theories have been proposed, but little evidence has been found to support these theories. The etiologies most commonly described for AFP and AO are:

1. Psychological
2. De-afferentiation, and
3. Vascular or neurovascular.

Of these theories, psychological disorders, usually depression is the most commonly mentioned one. Lesse (1956)^[16] described 18 patients with AFP and 8 patients with AO and he concluded that the pain complaints were entirely psychogenic in origin or represented a gross overreaction to a very minor organic deficit which had long since been considered. Melzack and Wall (1965)^[17] stated that the psychological factors are important in AFP, perhaps by opening or closing various "gates" either peripherally or centrally. De-afferentiation research has demonstrated that, after injury, organization and activity of central and peripheral nerves can change. This can result in chronic pain and other related symptoms (paresthesia, dysesthesia) for example, neuroma secondary to nerve trauma is thought to result in such pain. Rees and Harris (1978)^[9] and Sicuteri *et al.* (1991)^[18] suggested a vascular or neurovascular etiology. The patient can localize the area, even the tooth assumed to be the cause of the pain.

Maier and Hoffmeister (1989)^[19] stated that some aspects of AFP can be seen as a form of reflex sympathetic dystrophy. Both AFP and complex regional pain syndrome share common features, such as the disproportionate development after an initiating noxious event of relatively low intensity, the pain relief after sympatholytic intervention. An undefined patient-specific susceptibility factor, predisposing this population to a chronic pain syndrome, appears to be present. For decades,

emotional factors have been claimed to be the cause of chronic facial pain. Wilson (1932)^[20] reported seven cases of AFP in which he felt that emotional disturbances and the change in the behavior are out of proportion to the symptoms. Engel (1951)^[21] described AFP as a hysterical conversion symptom and was quite emphatic that the emotional disturbance was the cause. Feinmann *et al.* (1984)^[22] reported the possibility of predisposing and/or perpetuating affective disorders, somatization, personality disturbances or sleep disorders. Mock *et al.* (1985)^[23] said that a dental etiology must often suspected and indeed must be ruled first, but this can also result in extensive, usually unnecessary dental treatment, particularly endodontic therapy and extractions. Unfortunately, inappropriate dental treatment can lead to the perpetuation or even aggravation of the patient's pain. In some cases, the pain is precipitated by a dental or surgical procedure, or facial trauma, often quite minor. Other mechanisms involved in the pathogenesis of pain include sensitization of pain fibers, sprouting of adjacent afferent fibers, sympathetic activation of afferents, cross-activation of afferents, loss of inhibitory mechanisms and phenotypic switching of afferent neurons. These processes may underlie the clinical manifestation of AFP and AO.

Clinical Features

Sharav (1994)^[24] said that AFP is more common in women with an average between 40 and 50 years, but can range from 20 to 70 years, with a bimodal distribution. Although it is mostly found on one side of the face, a bilateral occurrence is not uncommon. Mock *et al.* (1985)^[23] said that the symptoms of AFP are initiated by a local, often surgical or trauma. In most cases, the trauma, which is not necessarily associated with nerve lesions, is relatively mild. Sessle (1992)^[25] suggested that de-afferentiation resulted in central neuronal hyperactivity in cases of nerve injury or trauma precipitated the symptoms.

According to Paulson (1997)^[26] AFP is characterized by an intense, deep and constant pain. The pain is burning or aching and is poorly localized. In general, the pain distribution does not follow anatomic pathways of the peripheral nerves. Pfaffenrath *et al.* (1993)^[27] reported allodynia, dysesthesia and paresthesia; such as feelings of tenderness, sensation of warmth, tingling or numbness as the sensory changes is common additional complaints of pain. Loeser (1994)^[28] and Bell (1989)^[29] stated that this is in contrast to trigeminal neuralgia, where the pain is well-localized, lancinating and paroxysmal with a defined trigger zone or site and unlike trigeminal neuralgia, eating, talking and other facial functions are usually not impaired in patients with AFP. In addition, the majority of those with AFP has no or minor limitations in their ability to work. Some patients have a history of dental treatment before the onset of pain; therefore cases may overlap with AO.

The molars and premolars in the maxilla are most often affected in patients with AO. Marbach (1978)^[7] said that AO presents as prolonged periods of constant throbbing or burning pain in teeth or the alveolar process and is usually characterized

by persistent toothache following pulp extirpations, apicoectomy or tooth extraction. This is in the absence of any identifiable odontogenic etiology observed clinically or radiographically. He also said that the pain in AO is chronic; however, the patients sleep is undisturbed, and there may be a brief symptom-free period on waking. Patients often have difficulty localizing the pain. Marbach (1978)^[7] and Lilly and Law (1997)^[30] reported that the site of the original trauma is the worst site for pain, but can spread to adjacent areas, unilaterally or bilaterally. He also said that the patients often seek multiple endodontic or surgical treatments, realizing no relief or even exacerbation of their symptoms.

It is likely that the AFP represents not a single clinical entity but rather several disorders, each with different etiological factors. Some may be true neuropathic pains, related to a peripheral nerve injury that cannot be documented or inappropriate activity within the sympathetic nervous system, such as has been suggested elsewhere in the body, and others may result a central nervous system disorder.

Differential diagnosis

Differential diagnosis of neuropathic pain conditions is the most challenging aspect of managing referred pain cases. Pain in the head and neck can be diverse. However, there are characteristics of odontogenic and neuropathic conditions that aid diagnosis. Furthermore, although there is some overlap in clinical presentation, careful examination of symptoms can differentiate trigeminal neuralgia from AO. The fact that neuropathic tooth pain can present exclusively intra-orally in the absence of obvious infection or trauma can be confusing to both patients and clinicians. Patients in dental environments are more likely to be considered to have dental pain as opposed to patients referred to a neurologist. This is where patients' perception of their problem can influence treatment and referral considerations. Careful history, clinical and radiological examinations are important.

Treatment

Once the diagnosis has been made, and other pathologies have been eliminated, it is important that the symptoms are taken seriously and are not dismissed as imaginary. Patients should be counseled regarding the nature of the pain and reassured that they do not have an undetected life-threatening disease and that they can be helped without invasive procedures. Loeser (1994)^[28] said that even small neurodestructive interventions carried out for therapeutic purposes generally lead to a worsening of this pain condition. When indicated, consultation with other specialists such as otolaryngologists, neurologists, or psychiatrists may be helpful. Mock *et al.* (1985)^[23] stated that patients usually have had many different treatment approaches leading to no relief or even worsening of the symptoms. Solomon and Lipton (1990)^[10] stated that may offer relief. Tricyclic amines such as amitriptyline, nortriptyline, and doxepin, alone or in association with phenothiazines, have been prescribed with

good results. Undesirable side effects require that tricyclic amines be titrated to the lowest clinically effective dose and discontinued if pain symptoms subside. Monoamine oxidase inhibitors have shown to be therapeutically successful in some specific cases. Some clinicians report benefit from topical desensitization with capsaicin, topical anesthetics, or topical doxepin. Analgesics and non-steroidal anti-inflammatory drugs either are ineffective or, at best, give temporary relief. Benzodiazepines (e.g. combination of chlordiazepoxide and amitriptyline) can be helpful in selected subgroups of AFP, such as in patients with a burning mouth syndrome. Schwartz *et al.* (1996)^[31] said that these patients are often angry about their treatment history and have a peculiar response to placebos or active drugs (e.g. tricyclics or calcitonin).

Maier and Hoffmeister (1989)^[19] said that the additional treatment options include transcutaneous electrical nerve stimulation, sympathetic nerve blocks, psychotherapy and behavioral approaches.

Conclusion

Pain is a universal phenomenon. As a dentist one comes across a number of patients who suffer from one form of pain or the other. Establishing the correct diagnosis is essential for successfully managing the pain condition. The management of orofacial pain is certainly a challenge to the clinician. Hence, the dentist has a great responsibility for the proper management of pain in and around the face, oral cavity and neck. The difficulty arises from the complexity of the many structures that make up the orofacial region. The dentist must differentiate between pain that are from dental, oral, and masticatory sources and those which emanate elsewhere. The dentist must also be able to identify complaints manageable on a dental level with dental techniques and methods. Identify complaints that, although related to oral and masticatory functioning, stem from causes that cannot be reasonably resolved with ordinary dental procedures and refer to a specialist.

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