

A Unified Concept of Idiopathic Orofacial Pain: Clinical Features

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The main features of atypical facial pain, stomatodynia, atypical odontalgia, and masticatory muscle and temporomandibular joint (TMJ) disorders are compared in this article, which included a search of articles indexed in MEDLINE. The fact that their terminology has been the subject of many debates can be considered a consequence of taxonomic difficulties and uncertainties. Epidemiologic studies indicate marked female predominance for all types of idiopathic orofacial pain. There is also a difference in the age of maximal prevalence between masticatory muscle and TMJ disorders and the other entities. The clinical presentations display several symptoms in common. Pain is oral, perioral, or facial and does not follow a nervous pathway. It has been present for the last 4 to 6 months or has returned periodically in the same form over a period of several months or years. The pain is continuous, has no major paroxysmal character, and is present throughout all or part of the day. It is generally absent during sleep. Clinical, radiographic, or laboratory examination does not reveal any obvious organic cause of pain. There is also a frequent presence of certain psychologic factors, personality traits, or life events. Based on these shared characteristics, a unified concept is proposed. Each of these entities belongs to a group of idiopathic orofacial pain and could be expressed in either the jaws, the buccal mucosa, the teeth, the masticatory muscles, or the TMJ.

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One of the most important trends in recent research in temporomandibular disorders has been to attempt to define accurately the different disease processes and to standardize a classification scheme. Much clinical research has been undertaken to define each subgroup of masticatory muscle and temporomandibular joint (TMJ) disorders. This approach has led to a new classification of 12 disease processes, which form the group of masticatory muscle and TMJ disorders. Each may be present independently or simultaneously in a single patient. Diagnostic criteria for each subgroup of this classification have been developed and published, with the primary goal to standardize research efforts in this area.^{1,2} As a result, knowledge of pain due to masticatory muscle and TMJ disorders has greatly increased, but many related orofacial pain syndromes have been left unclassified. It is possible that an equal amount of knowledge could be gained by looking beyond the masticatory muscle and TMJ disorders to a definition of the other types of idiopathic orofacial pain.

Atypical facial pain, atypical odontalgia, stomatodynia, and masticatory muscle and TMJ pain disorders display common clinical characteristics. Partly for this reason, a unified concept of idiopathic orofacial pain has already been suggested.^{3,4} In addition, several of the proposed etiologic mechanisms could also be shared; for example, alteration in the balance of female steroid hormones, modification by lesion and/or sensitization of nerve fibers, or involvement of the sympathetic nervous system or psychologic factors, such as life events. Evidently, clinical and pathophysiologic differences exist, but they could result from the obvious but possibly misleading fact that they arise from 5 different tissues: bone, tooth, oral mucosa, TMJ, and masticatory muscles. It can be postulated that a unique disease would be differently expressed in different tissues. Recently, the priority given to anatomic criteria in the current pain classification systems⁵⁻⁷ has been questioned⁸ because this may reclassify pain entities that share common clinical features and/or mechanisms purely on an organ or tissue basis. It is useful therefore to review the clinical features and mechanisms of these types of persistent orofacial pain with regard to classification for purposes of both research and treatment.

This article aims to bring together the terminology, epidemiology, and signs and symptoms for each of the idiopathic orofacial pain subgroups and to propose diagnostic criteria that will allow for comparison between each entity.

Atypical Facial Pain

Definition and Terminology

Despite the position taken by both the International Association for the Study of Pain (IASP)⁵ and the International Headache Society (IHS),⁶ the term "atypical facial pain" continues to be used by almost all authors. There is no definition for atypical facial pain, precisely because the term was coined to cover all unexplained cases of pain. In the literature this term is used to describe 2 totally different concepts. The first is that described by the IASP⁵ and the IHS,^{6,9} which suggests that the term should be abandoned in favor of "other and unspecified pain in the jaws" or by "facial pain not fulfilling other criteria." These terms would regroup all the intermediate clinical situations that do not fall into one of the well-defined categories, such as cluster headache, trigeminal neuralgia, neuropathic pain associated

with systemic disease or trauma, and several types of migraines or tension-type headaches. The term is thus employed as a "wastebasket" definition, which can only be applied by elimination. What is more, certain other subgroups that had not been identified at the time, such as TMJ disorders or cluster headache, could also be included in this group.¹⁰ The second diagnostic concept of atypical facial pain is of that of a defined set of characteristics that aim to describe a relatively homogeneous subgroup of facial pain. Diagnosis thus becomes a positive procedure rather than one accomplished by elimination. This article will consider atypical facial pain as a distinct group.

Atypical facial pain can be described as a chronic pain of unknown etiology that is felt continuously throughout all or part of the day within the bone or deep tissues of the orofacial region. Other expressions that are no longer used as synonyms for atypical facial pain include atypical facial neuralgia,¹¹ dental causalgia,¹² or phantom orofacial pain.¹³

Epidemiology and Demography

No evaluation has been made of the prevalence of atypical facial pain in the general population. Existing figures are drawn from clinical impressions or from studies of clinical populations. Its prevalence would seem to fall between that for trigeminal neuralgia, which is very low, and that for masticatory muscle and TMJ disorders, which is high. All the studies¹⁴⁻¹⁸ indicate a very high preponderance of female sufferers in these populations—between 3 and 10 females for every male affected. This may be the result of a truly high prevalence among women or perhaps a more frequently expressed treatment need in the female population. It is possible that men may be more readily able to assimilate the pain into their way of life (see references in Fillingim and Maixner¹⁹). The average age affected is that of the postmenopausal period or above. The reported average age on consultation was 52 years, with extremes of 24 and 82 years.^{16,18} The average age of onset was 45 years.¹⁵ Thus, atypical facial pain concerns chiefly menopausal or older women.

Description

As the definition of atypical facial pain was originally made only by elimination, its characteristics were first described in comparison with those of trigeminal neuralgia.^{15,20-22} Both are marked by extremely intense pain that is usually localized to

the mouth, jaws, or face. In reality, the pain characteristics are not comparable, and a differential diagnosis between them is rarely a problem.²³ The pain location, for example, is different. Atypical facial pain often is of a fluctuant nature, and it is felt deep within the tissues,²⁴ in the bone of the maxilla or mandible. Trigeminal neuralgia is most often felt in the superficial tissues only and always in the same locus.

The timing of episodes of pain is also different. Although in both cases the pain is experienced only while the patient is awake, it is continuous in atypical facial pain.²¹ In atypical facial pain there is no true trigger zone, although the pain may be intensified, or more rarely decreased, by functional movement (for example by mastication or speech). Episodes of increased pain are never paroxysmal, as they are for trigeminal neuralgia or certain other neuropathic pains.²⁵

At the onset of the disease, pain is often felt within a defined zone of the oral cavity or midface that has undergone some sort of trauma. The trauma may be accidental or therapeutic in nature (eg, endodontic treatment, dental extraction, or sinus surgery)¹⁸ and the initial recovery may be difficult.¹⁴ The pain is described either as having been aggravated by the injury or as having been initiated by it.^{14,15} Sometimes the pain reappears only after a period of months or years following the original pain experience. With time, the pain spreads out over a larger territory. It does not respect any particular nervous pathway. In one third of cases, pain becomes bilateral.^{16,21} Often many different dental or surgical treatments are attempted, with no results other than an increase in symptoms¹⁸ and partial or total edentulousness.¹⁴

Pain intensity is described as moderate to intense and is equivalent to that described for trigeminal neuralgia. Recently experienced pain is described as the worst possible pain 3 or 4 times more often than that of pain of masticatory muscles and TMJ disorders.²⁶ To describe the qualitative aspect of their pain, patients often use the emotional terms of the McGill Pain Questionnaire,²⁰ such as "vicious" or "excruciating." Strangely, they often evoke mechanical descriptors such as "tugging," "constriction," "tightening," "piercing," or "movement" inside the underlying bone.¹⁶ The pain is also often described as "burning."¹⁶

Neurologic signs are not obvious, but there may be dysesthesia, allodynia, and/or paresthesia.^{14,16,21} Paresthesia is described in diverse terms, such as "numbness" or "pins and needles."^{16,21} Signs and symptoms due to sympathetic or vascular

dysfunction may also be present. Subjective impressions of warmth or swelling of the mucosa or soft tissues are frequent and are sometimes confirmed clinically by the presence of erythema or edema.²⁵ These signs are usually discrete, and they may be constant, or more often, undergo periods of spontaneous remission. Change, or an impression of change, in salivary flow may also be noted. Recently, an area over the apices of the last 2 maxillary molars has been described as being hypersensitive to pressure and has been shown to display a temperature rise.^{27,28} Analgesics give little or no relief. A transient but complete relief follows the administration of local anesthetic to the affected edentulous area.¹²

A large proportion of patients report associated general symptoms, such as chronic, cervical, or lumbar spinal pain; migraines; cutaneous pruritis; irritable bowel syndrome; or dysmenorrhea.³

Psychologic signs are acknowledged by all authors, but debate surrounds the type of pathology concerned. Depression, anxiety, intense stress, or a distressing life event in the 6 months preceding the onset of pain have all been implicated.^{3,4,16,24} It is also important to compare the incidence of psychologic problems within this population and that of the general public (see references in Feinmann and Harris²⁹). Most important, the existence of a causal link between psychologic factors and the disease is highly contentious.^{3,30} It should also be noted that these patients are often cancerophobic.

These patients express a high level of demand for invasive treatment,¹⁸ which leads to the consultation of many health care professionals. Pfaffenrath et al showed that patients had consulted an average of 7.5 professionals, in the following descending order: dentist; general medical practitioner; neurologist; ear, nose and throat surgeon; orthognathic and maxillofacial surgeon; psychiatrist; ophthalmologist; and dermatologist.¹⁶ The edentulous region, which is so often the result of these consultations, then presents a functional problem for the patient, who is unable to cope with removable prostheses and who complains of problems with chewing. The impact on other aspects of quality of life has not been studied, but it seems that capacity to work is only relatively slightly decreased.¹⁶

Diagnostic Criteria

The first diagnostic criteria that were proposed in the literature probably lack specificity.¹⁴ The criteria suggested by the IHS^{6,9} have been questioned

by Pfaffenrath et al,¹⁶ who noted that if these criteria are applied strictly then they must lead to a number of false negatives,¹⁶ and they therefore proposed certain changes to improve the rate of inclusion and the sensitivity. Unfortunately, since the choice of their population of sufferers of atypical facial pain is not fully described, it is difficult to judge the validity of their conclusions. The proposed criteria for atypical facial pain, presented here, are derived from those of the IHS.^{6,9} They have not been validated.

- The pain is initially well-circumscribed and is limited to a zone of the face or oral cavity. The pain may then spread over a larger area and become diffuse. It is experienced deep within the tissues.
- The pain has been present for 4 to 6 months or more (remission is sometimes possible).
- Pain is continuous throughout all or almost all the day except during sleep.
- The pain does not have a major paroxysmal component.
- There is no definitive etiology, and a diagnosis cannot be confirmed by any one clinical, radiologic, or laboratory examination.
- There is no associated hypoesthesia.

The IHS adds that the pain may be initiated by a surgical intervention or trauma to the face, teeth, or oral mucosa. There is a high female predominance.

Atypical Odontalgia

Definition and Terminology

Atypical odontalgia may be defined as pain of dental origin without a definitive organic cause. Forms of oral pain that are not of dental origin must be eliminated. Atypical odontalgia probably represents a clinical form of atypical facial pain^{25,31,32} whose separate classification is justified by its localization to 1 or more teeth and by the precise description of its characteristics that has been given by Graff-Radford and Solberg.³³ The term "phantom tooth pain" has been proposed to describe a similar phenomenon that is intermediate between or that resembles both atypical facial pain and atypical odontalgia.^{34,35} Other terms have been used that describe a disease approaching the clinical picture of atypical odontalgia or that of atypical facial pain. These include "idiopathic odontalgia," "idiopathic periodontalgia," "vascular toothache," "migrainous neuralgia,"

"neurovascular odontalgia,"^{33,36,37} and more recently "neuropathic orofacial pain."³⁸

Epidemiology and Demography

The prevalence of atypical odontalgia in the general population is unknown. However, knowledge of the frequent association between endodontic treatment and the onset of pain inspired 2 retrospective studies^{39,40} of patients who had undergone endodontic treatment. At least 8 patients in the first study and 3 in the second study presented with atypical odontalgia; this represented a minimal prevalence of 2.5% and 3%, respectively, for the 2 populations studied.^{39,40} Recent figures give a slightly higher estimate.⁴¹ Women constituted 68 to 100% of the populations studied,^{5,31,32,37,42,43} with an average age between 40 and 51 years.^{33,37,42,43}

Description

Pain is localized to a tooth that shows no discernible pathology to cause such symptoms. Premolars and molars are most often affected, and those in the maxilla are more often affected than those in the mandible.³³ The term "phantom tooth"^{34,40} refers to the situation when a tooth that has been extracted seems to be the source of pain. It is difficult to tell whether the pain is experienced in the dental tissues or in the bone that has replaced the roots of the tooth. Pain is continuous throughout the day, or part of it. Sleep is not disturbed.³⁴ Symptoms may be present over several months or may come and go periodically over the years, but the pain is always of the same nature.³⁴ The tooth or teeth concerned may be hypersensitive to thermal stimulation but not to percussion. The overlying alveolar bone may be extremely sensitive to moderate, sustained pressure.³³ The pain is moderate to intense⁴³ and presents little³⁴ or no³³ paroxysmal characteristic. Description of the pain is very varied⁴³ but is often "throbbing" or "aching."³³

Symptoms begin in adulthood and are reported by authors to be usually^{34,43,44} or always³³ initiated by dental treatment, although the treatment may be as noninvasive as the preparation of an occlusal rest.³³ The patient often demands treatment that leads to root treatments and to extractions, which are all the worse for the fact that they tend to increase the intensity and the spatial extent of the pain suffered.³¹ Following extractions, the pain may be transferred to a tooth adjacent to the edentulous space.⁴⁰ The center of the pain is often

localized over the neck of the tooth. It is possible that following successive extractions, the course of the atypical odontalgia becomes atypical orofacial pain.

The success of local or regional anesthesia is equivocal.³⁷ Anesthesia may be achieved without a change in the intensity of pain, which would suggest a neuropathic origin. This sign has been proposed by Graff-Radford and Solberg as a diagnostic criterion.^{33,45} Allodynia is frequently experienced.^{33,34}

The link between atypical odontalgia and psychologic signs is even more strongly disputed than for the other subgroups of atypical facial pain. The few discrete psychologic problems observed in a large number of cases may be explained by the nature of the pain itself.⁴⁶ The possibility of a predisposing psychologic factor is, however, considered.^{3,32,40,46}

Diagnostic Criteria

The criteria presented here are similar to those proposed by Graff-Radford and Solberg.³³ They have not been validated.

- Pain is localized to a tooth that is present in the mouth or has recently been extracted.
- Pain has been present for the last 4 to 6 months or has returned periodically in the same form over the last period of months or years.
- Pain is continuous throughout all or part of the day except during sleep.
- The pain has no major paroxysmal character.
- Clinical or radiographic examination does not reveal any obvious cause of pain.

Other factors that are often associated:

- Allodynia (termed hyperesthesia by Graff-Radford and Solberg³³)
- Unreliable effect of local anesthesia

Stomatodynia

Definition and Terminology

Stomatodynia is characterized by pain in the oral mucosa that cannot be attributed to any known organic cause. This definition excludes pain from the lingual mucosa or from the oral mucosa that could be explained by local or systemic pathology. When pain or burning of the mucosa are caused by a known disease process, it reflects only 1 symptom of this pathology and cannot be classed

as a separate entity. When stomatodynia is defined as above, then it can be classified apart and not merely as a symptom. This explains why the authors prefer this definition to that of the IASP,⁵ "burning pain of the tongue or mucosa," which describes a symptom. The term "stomatodynia" is preferable to that of "burning mouth syndrome" for the same reasons. Many other terms have been proposed that emphasize one aspect or another of the disease, eg, glossodynia, idiopathic glossodynia, sore mouth, burning tongue, oral dysesthesia, stomatopyresis, or glosopyresis.⁴⁷⁻⁴⁹

Epidemiology and Demography

An important variable between the epidemiologic studies presented in the literature is the choice of the sample, which is rarely representative of the general population. This is emphasized in a study in which 3 different populations were sampled for comparison. It was found that the prevalence of oral pain among patients attending a menopause, diabetic, or general dental clinic was 26%, 10%, and 2.6%, respectively.⁵⁰ Another difficulty arises from the imprecision of the definitions used to reach the diagnosis of stomatodynia. A very high prevalence may be reported by studies that take into account all forms of pain or burning sensation in the oral mucosa. This is particularly true for epidemiologic studies undertaken with postal questionnaires.^{13,50-52} These factors could explain why a prevalence as high as 15% has been reported in the general population.⁵¹ The studies of the Toronto group demonstrate this well. They found an initial prevalence of 4.5% among the general population of Toronto following a postal questionnaire.⁵³ When these results were followed up by a telephone interview, a more precise diagnosis was made, and the prevalence fell to 1.5%.⁵⁴ Even according to the authors this figure is probably unrealistically high.⁵⁵ The National Health Interview Survey is a carefully developed study that was applied to the whole of the United States. It found 0.7% positive replies to the question "During the past 6 months, did you have more than 1 prolonged, unexplained burning sensation in your tongue or any other part of your mouth?"¹³ Since a positive reply to this question does not give a definite diagnosis of stomatodynia, it can be assumed that the true prevalence is in fact below 0.7%. This figure still seems high, and the fact that another study found a totally different estimate (less than 0.01%)⁴⁷ suggests that current data are unreliable.

Women are much more frequently affected than men. The relative proportion is between 3 and 20 females for each male, depending on the study.^{48,49,54,56-59} These figures may be biased by the fact that women may be more likely than men to seek medical assistance.¹³ Women affected are menopausal or postmenopausal^{48,49,56,57,59-62} and have an average age of approximately 60 years.^{57-59,61}

Description

Many descriptions can be found in the medical literature.^{5,48,49,51,54-58,63,64} The patient describes pain, or occasionally dysesthesia, localized to the buccopharyngeal mucosa, that shows no organic sign of pathology. The most frequently affected areas are the tongue, the palate and gingivae, the lips, and the pharynx.^{56,58,65} Pain is generally bilateral and symmetrical⁵⁸ and is always independent of a nervous pathway.

Pain is continuous throughout all or part of the day and tends to worsen over the daytime.^{57,58} It usually occurs daily.³⁶ Sleep disturbance may be recorded as a parallel phenomenon,⁵⁸ but there is not a causal relationship, as loss of sleep is rarely due to the presence of pain.^{57,66} Stomatodynia is generally present over a number of years^{54,67} but there may be periods of remission.⁵⁴

Pain is generally spontaneous, but in certain patients it may be triggered by certain foods, particularly spicy or acidic foods.^{57,58,65} In other patients, food or drink may alleviate pain.^{57,65} Other daily activities may alter the intensity of the pain sensation.⁵⁷ Pain intensity varies greatly between patients, ranging from a simple irritation to the worst pain imaginable. There is no major paroxysmal component. The usual term found to describe the pain is "burning."⁶⁵

Subjective impressions of xerostomia (dry mouth),^{57,61,65} thirst,⁵⁷ or dysgeusia (alteration in taste perception)^{55,57,58,65,68} often accompany the pain. Certain psychological disorders are often noted, most frequently depression or anxiety.^{58,65-67,69} Rojo et al⁶⁹ found that their patients with stomatodynia were divided into 2 different groups of equal size. The first presented more symptoms of anxiety, depression, obsession, somatization, and hostility than the control group, while the second group was not distinguishable from the control. This shows that although anxiety and depression are frequently encountered in this population, they are by no means present in all cases.^{65,69} In addition, many patients with stomatodynia are cancerphobic.^{48,49,57}

Reported changes in diet linked to this disease could be a result of the aggravation of pain intensity that can be experienced with certain foods in certain patients.⁵⁷

Physiopathogenic Hypotheses Limited to Stomatodynia

The fact that the disease affects primarily postmenopausal women suggests that hormonal or degenerative factors may be implicated in the etiology of the disease. It is, however, essential to exclude burning sensation of the oral mucosa due to nonspecific causes, such as the sequelae of radiotherapy, mucositis associated with chemotherapy, or xerostomia induced by psychotropic medication. Other factors that can cause pain identical to that of stomatodynia are iron deficiency anemia or *Candida albicans* infection, but these will not be considered here. It seems that these causes of burning oral pain occur less frequently than burning pain of idiopathic origin.⁵⁷ Other specific local etiologic factors that have been proposed for stomatodynia include allergy, electrogalvanism, presence of a partial denture, parafunction, or salivary gland dysfunction. None of these proposals have been scientifically confirmed.^{49,54,55,63,70,71} Similarly, certain systemic disorders have been evoked but their implication never proven.⁶² These include Gougeraud-Sjögren syndrome, diabetes, and vitamin deficiency, but any link is unlikely to be causal.^{49,54,55}

Diagnostic Criteria

The criteria proposed here are not validated and exclude oral pain that is a result of defined local or systemic disease.

- Pain or dysesthesia in the buccopharyngeal mucosa.
- Pain has been present for the last 4 to 6 months or has returned periodically in the same form over the last period of months or years.
- Pain is continuous throughout all or part of the day except during sleep.
- There is no major paroxysmal character.
- Clinical or radiographic examination does not reveal any obvious cause of pain.

The following characteristics are noted:

- High female prevalence
- Presence of a depressive, anxious, or somatic psychological factor

Idiopathic Facial Arthromyalgia

Many original articles and reviews have been published on masticatory muscle and TMJ disorders. It is out of the scope of this paper to exhaustively review all the classifications, epidemiologic data, clinical features, and proposals for diagnostic criteria. The reader is referred to the relevant reviews.^{2,7,72-77} The present chapter is aimed at presenting the arguments in favor of inclusion of what are generally called the temporomandibular disorders within the general concept of idiopathic facial pain.

Definition and Terminology

"Temporomandibular disorders" is the internationally established term^{74,75} for these conditions. Up to this point they have been called "masticatory muscle and temporomandibular joint disorders." These are collective terms that embrace a number of clinical problems involving the muscles of mastication, the TMJ, and associated structures.^{7,73,78,79} The TMJ disorders are considered a subclass of musculoskeletal disorders,^{73,79} and the similarities between some of their pathophysiological features with other diseases, such as fibromyalgia, tension headache, or low back pain, have recently been stressed.⁸⁰

In the past, TMJ disorders were considered a single homogeneous syndrome. This belief can be seen in the many different terms that were used, such as Costen syndrome,⁸¹ TMJ dysfunction,⁸² pain dysfunction syndrome,^{83,84} or myofascial pain-dysfunction syndrome.⁸⁵ Modern thinking supports the view that the TMJ disorders represent a cluster of related diseases that have many clinical features in common and whose signs and symptoms are localized to the TMJ and/or muscles of mastication.^{7,78} As a consequence, a large amount of work has been dedicated to the division of this group into several subgroups whose signs and symptoms overlap.¹ In this classification system, multiple diagnoses are possible, meaning that a single patient can be categorized into several different divisions. This approach has proved useful, as it has deepened knowledge of the clinical subgroups of TMJ disorders and has given clinicians a better basis from which to choose a suitable treatment plan. It has also given the researcher a nominally homogeneous group of patients. This classification may, however, be misleading from a taxonomic and pathophysiological point of view. No data indicates that different mechanisms are acting in each of the different subgroups. As Turk

stated⁸⁶: "There is no way of knowing whether the characteristics that constitute the proposed categories reported actually exist or are artificial constructions based primarily on clinical experience." Moreover, the extensive overlap of signs and symptoms could be more than a "disturbing trap for scientists and clinicians" and could indicate that patients are scattered in a continuum, with no isolated group of patients corresponding to a clinically well-defined illness.

Another point of view is to distinguish 2 groups of patients within the category of TMJ disorders. The first group consists of patients suffering from pain with an identified somatic cause, either related to general disease, such as neoplasia or rheumatoid arthritis, or to a degenerative TMJ problem. The second group, much larger, consists of patients suffering from pain that cannot be easily explained by a somatic origin, at least with the presently available knowledge. In this latter case, unexplained pain related to the masticatory muscles and/or TMJ is the main sign, and we propose, after Feinmann et al,^{24,29} to term these conditions "idiopathic facial arthromyalgia." The term "dysfunction" can easily be discarded given the fact that almost all patients (97%) seek treatment because of pain.⁸⁷

Many recent systems of classification differentiate between 3 groups of TMJ disorders, ie, myofascial pain, intracapsular or disc disorders, and degenerative disorders (see Clark et al⁷⁷). It is tempting to distinguish idiopathic facial myalgia from idiopathic facial arthralgia, the former corresponding to the facial form of myofascial pain syndrome and the latter to the disc derangement disorders. There are several reasons not to do so. True idiopathic facial myalgia syndrome frequently presents without joint signs. In this group, therefore, pain is the principal feature and appears as the first diagnostic criterion, even if other usual signs of TMJ disorders are present.⁷ On the other hand, the leading clinical feature of disc derangement disorders is related to the dysfunctional joint and not to the pain itself. In the case of a pure disc disorder without pain, the need for treatment should be carefully considered, since aggravation of the condition is uncommon.^{7,88,89} In addition, there is now much data that clearly shows a poor correlation between disc dysfunction and noises from the joint,⁹⁰⁻⁹² which are commonly sought during clinical examination as a basic sign for diagnosis. Therefore the primary indication for treatment of the disc derangement disorders is joint pain rather than dysfunction, the former being also frequently associated with muscle pain.

Table 1 Summary of the Principal Signs and Symptoms of the Different Types of Idiopathic Orofacial Pain*

Sign/symptom	Atypical facial pain (bone)	Atypical odontalgia (tooth)	Stomatodynia (mucosa)	Idiopathic facial arthralgia (muscle, articulation)
Localization	Independent of a nervous pathway. Unilateral initially, becomes bilateral ^{16,21,30}	Independent of a nervous pathway. A single tooth initially but may spread ³	Independent of a nervous pathway. Often bilateral and symmetrical ⁵⁸	Independent of a nervous pathway. Unilateral or bilateral
Time period of pain	Continuous ^{21,25}	Continuous with possible remission ³⁴	Continuous	Continuous with remissions
Paroxysmal	No	No ³³ or little ³⁴	No	No
Pain during sleep	No ^{3,30}	No ^{3,34}	Infrequent ^{5,57,107}	Uncommon ¹⁰⁴ but disturbed sleep ¹⁰⁵
Intensity	Moderate to intense ²⁵	Moderate to intense ⁴³	Weak to intense ⁶⁵	Weak to intense
Descriptors	Emotional, ²⁰ mechanical, ¹⁶ burning ^{16,108}	Varied ⁴³	Burning ⁶⁵	Spontaneous (dull, aching) or during function or voluntary movements
Initiating factor	Frequently trauma ^{15,18}	Frequently dental caries ^{34,43,44}	Stressful life event may be implicated ^{65,70}	Possible direct trauma ¹⁰⁹ , whiplash, bruxism possible
Effect of local anesthetics	At least temporary relief ¹²	Ambiguous effect ^{33,37,45}	No data	Temporary relief (for trigger points)
Neurologic signs	Frequently dysesthesia, allodynia, paresthesia ^{14,16,21}	Frequently allodynia ^{33,34}	Infraclincal ^{55,110,111}	Allodynia (trigger points in myofascial pain)
Effect of analgesics	None or weak ³	None or weak ³	None or weak ³	Sometimes effective
Psychologic factors	Frequent ^{3-5,9,16,108}	Debatable ^{3,32,40,46}	Frequent ^{6,58,65-67,107}	Frequent
Sympathetic signs or implication	Sometimes ^{25,27,28}	No but possibly implicated ⁴³	Not described	Implication suggested for myofascial pain
Other associated local signs	Bone cavity, ^{44,112-117} osteoporosis?	None	Xerostomia, ^{57,61,65} dysgeusia, ^{50,55,57,58,68} thirst ⁵⁷	Limited range of mandibular motion, masticatory and TMJ palpation tenderness, TMJ sounds (non-specific), ¹¹⁸ bruxism and oral habits
Mean no. of previous consultations	7.5 ¹⁶	5 to 6.2 ^{33,43}	2.5 ¹¹⁹	3.2 ¹²⁰
Prevalence	Unknown	2.5 to 3% of endodontic cases ^{39,40}	< 0.7% ^{13,47}	4 to 5% ⁹⁷
Mean age (y)	52 ^{16,18}	40 to 51 ^{33,37,42,43}	55 to 60 ^{57-59,61}	Second to fourth decades of age ⁷⁹
Male:female ratio	1:3 to 1:10 ¹⁴⁻¹⁸	1:2 to 1:20 ^{31-33,37,42,43}	1:3 to 1:20 ^{48,49,54,56-59}	1:3 to 1:9

*The description of idiopathic arthralgia is that typically used for the TMJ disorders (from Okeson⁷ or Lipton and Dionne¹⁰² if not otherwise indicated), although the two terms do not cover exactly the same subgroups.

Therefore, this is in line with the more general concept of arthralgia of the face (Table 1).

Epidemiology and Demography

There are many sources of confusion in the epidemiologic studies that have examined the prevalence of masticatory muscle and TMJ disorders. The wide variety of clinical presentations and the absence of objective diagnostic criteria have driven a research group to define both a system of classi-

fication it allows for multiple diagnoses and a list of Research Diagnostic Criteria, which are intended to fit validation processes.^{1,2}

Epidemiologic studies should also take the natural history of the disease into account. Longitudinal studies have shown that the semiology of this group of diseases varies with age in a non-random fashion. Marked variation is noted in the intensity of pain and in the presenting of signs and symptoms over time.^{93,94} The prevalence of idiopathic arthralgia is greatest among young

adults and lessens from 40 years of age.⁷⁹ The traditional belief in an inevitable aggravation of problems is not valid. When the results of epidemiologic studies are interpreted, the difference must also be noted between an isolated or weak sign and intense, lasting pain that induces the patient to seek treatment.

Despite these reservations, the prevalence of masticatory muscle and TMJ disorders has begun to be better recognized and can be summarized as follows. Presence of an isolated sign is extremely common, with 76% of a sample of North American students presenting with at least 1 sign, males and females alike.⁹⁵ A recent study⁹⁶ reported a prevalence of at least 1 objective sign among 44% of subjects. This study was undertaken on a large representative sample of the Dutch population. The same group confirmed these figures but showed by meta-analysis that there is a great range of prevalence of signs (between 0 and 93%). The prevalence of pain is reported as being between 5 and 30%, depending on the criteria used for definition of intensity and frequency of symptoms. In North America, a consensus value of 12% has been established for pain suffered in the 6 months preceding study.⁹⁷⁻¹⁰⁰ Despite this, the need for treatment for this type of pain only approaches 4 or 5%, and only 2% of patients had sought treatment in the 9 months preceding study.⁹⁷

Of those patients seeking treatment, the majority are female (3 to 9 females for each male^{7,79,95}). The pain is of greater severity for women, both physical and psychologic, among those seeking treatment.¹⁰¹

Description

Only a very short description of the signs and symptoms based on the guidelines edited by the American Academy of Orofacial Pain (AAOP)⁷ and on the National Institutes of Health (NIH) technology assessment conference on management of temporomandibular disorders¹⁰² will be presented. Detailed descriptions of the different subgroups can be found in many books and reviews.^{7,73-76,102} A comparison of the signs and symptoms of idiopathic orofacial arthromyalgia with those of the other types of idiopathic orofacial pain is presented in Table 1.

Pain is by far the most frequent symptom of TMJ disorders⁸⁷ and is by definition the basic one when idiopathic facial arthromyalgia is considered. Pain is located in the muscles of mastication, preauricular area, or TMJ⁷⁹ on one or both sides.

It frequently radiates to other parts of the face but does not follow a nerve trajectory. Pain is continuous but can be triggered or exacerbated by movement or function. Intensity may vary over each 24-hour period¹⁰³ and is usually experienced only during the day,¹⁰⁴ even if patients often complain of disturbed sleep.¹⁰⁵ Variety also exists in the length of episodes of pain and remission between episodes. The pain is generally weak to moderate and is often described as dull or aching. There is no major paroxysmal character and few neurologic signs. In myofascial pain, however, some kind of allodynia is present, as trigger points can be identified.

Limitation or asymmetry of mandibular movement; noise in the joint described as clicking, popping, or crepitus; and association with bruxism are frequent. The specificity or the lack of specificity of these signs has been the subject of many recent studies.^{77,90-92} Some other nonspecific and unexplained complaints are tinnitus, ear fullness, and perceived hearing loss. Psychologic factors are frequently present and may predispose or perpetuate the condition.

Diagnostic Criteria for All Orofacial Pain Entities

Partly validated diagnostic criteria are available for the different subgroups of TMJ disorders.^{2,7,106} No criteria have been proposed for the different types of idiopathic arthromyalgia. The description of pain found in the literature does, however, allow diagnostic criteria common for the whole group of idiopathic orofacial pain to be proposed.

- Pain is oral, perioral, or facial and does not follow a nervous pathway.
- Pain has been present for the last 4 to 6 months or has returned periodically in the same form over a period of several months or years.
- Pain is continuous and is present throughout all or part of the day and is infrequent during sleep.
- There is no major paroxysmal character.
- Clinical, radiographic, or laboratory examination does not reveal any organic cause of pain.

The following characteristics are noted:

- A marked female predominance
- The frequent presence of certain psychologic factors, personality traits, or life events

Taxonomy and Research Perspectives

Classification of pain in the head and neck region into specific diseases, syndromes, or pain entities relies largely upon work undertaken by the IASP⁵ or by the IHS^{6,9} and later completed by the AAOP.⁷ Difficulty in classifying the subgroups of idiopathic facial pain entities is illustrated markedly in these reference texts. The 4 distinct groups of atypical orofacial pain are never grouped together and are not even always described individually. Furthermore, in the introduction of a classification for all types of pain, the experts of the IASP⁵ cite atypical facial pain under the title of "some controversial issues." For Merksey and Bogduk,⁵ the term "atypical facial pain" is excluded from the classification because it does not represent a well-defined entity and could correspond to different pathologic situations, depending on the case (such as TMJ disorders, atypical odontalgia, or migraine). In this classification, the 2 subgroups of muscle and joint pain (myofascial pain and arthralgia) are not separated.

The IHS,^{6,9} for its part, differentiates neither stomatodynia nor atypical odontalgia. Myofascial pain of the face is classified, but the myofascial subgroup is linked to tension-type headaches. The term "atypical facial pain" is also absent from this classification and is replaced by a group entitled "facial pain not corresponding to any of the preceding groups." This phrase is used interchangeably with the term "atypical facial pain" by the AAOP,⁷ which associates it with atypical odontalgia and with sympathetically maintained pain. Stomatodynia and the different groups of muscle and TMJ pain are described but are not linked together or with the other idiopathic facial pains.

This brief review of the best-known classifications emphasizes the confusion that reigns in this area. The principal criterion used in the classification of pain is that of localization.⁵ This principle can overlook the similarities between the subgroups of idiopathic orofacial pain, whose main differentiating factor is the tissue from which the pain is experienced. Table 1 underlines the common clinical characteristics and argues in favor of a concept of a group of diseases brought together under the heading "idiopathic orofacial pain." It also emphasizes the need for epidemiologic studies aimed at both the clarification of taxonomy and the validation of diagnostic criteria, which will allow the division of patients into homogeneous groups. Epidemiologic studies of large numbers of patients are needed to collect all the semiologic data. Then, cluster analysis of the distribution of

signs and symptoms should allow the definition of distinct entities on a scientific basis. Epidemiologic characteristics also need to be studied. While the prevalence, evolution without treatment, and the populations at risk are beginning to be elucidated for muscular and TMJ disorders, this is not the case for the other potential subgroups of idiopathic orofacial pain. In particular, the prevalence of atypical facial pain in the general population is totally unknown, despite its impact on patients' lives. Its prevalence among populations at risk, for example in menopausal women, would have important clinical implications. Also, it could be presumed that the prevalence of stomatodynia would be, in these groups, higher than the current estimate of less than 1% in the general population. Although longitudinal studies are not feasible, projects studying the natural history of atypical facial pain or stomatodynia would be clinically useful. This type of data could influence the politics of public health. Finally, a better knowledge of taxonomy and epidemiology of these diseases would be helpful in defining inclusion criteria of clinical trials aimed at the improvement of treatments.

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