Development and Validation of Classification Criteria for Idiopathic Orofacial Pain for Use in Population-based Studies

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Aim: *To develop and validate a questionnaire-based tool which would enable classification of idiopathic orofacial pain in the general population.* ***Methods:*** *A postal questionnaire-based cross-sectional survey was made of 4,200 randomly selected adults who were registered with a general medical practice in North West England. The questionnaire collected information on a number of factors: demographics (age, gender), orofacial pain (duration, descriptors, site, pattern, intensity, disability, and consultation behavior), and comorbidities (reporting of other unexplained symptoms and psychosocial factors). Subjects reporting orofacial pain were interviewed by an examiner blinded to their exposure status and classified into 1 of 3 categories:* (a) *dentoalveolar,* (b) *musculoligamentous/soft tissue, and* (c) *idiopathic orofacial pain.* ***Results:*** *A high adjusted response rate of 72% was achieved (crude response rate 60%). Of those who reported orofacial pain and were eligible for interview (n = 218), 197 (88%) were interviewed. Subjects classified by interview into the idiopathic category were more likely to report aching, nagging, and chronic pain pain at multiple sites. They were also more likely to report facial trauma and other chronic symptoms and to have consulted multiple health-care workers. Variables that most strongly predicted membership into the idiopathic category were female gender, nagging, aching pain which was worse when stressed, and topography (pain at multiple sites and unilateral pain).* ***Conclusion:*** *The classification criteria developed for idiopathic orofacial pain can be used as a screening tool for subjects with this condition in the general population.* J OROFAC PAIN 2007;21:203–215

Key words: classification, general population, idiopathic, orofacial pain

There are 4 recognizable, frequently-coexisting symptom complexes of chronic idiopathic orofacial pain: temporomandibular disorders or TMD (myofascial face pain); atypical facial pain (atypical facial neuralgia); atypical odontalgia (phantom tooth pain); and burning mouth (oral dysesthesia, glossodynia, glossopyrosis). These conditions may be considered medically unexplained symptoms affecting 4 regions of the mouth and face,[1,2] although some researchers choose to treat them as separate entities.[3] Evidence from a recent study, albeit in a selected sample in tertiary care,[4] has shown that these idiopathic facial pain entities tend to cluster together into a single group.

The vast majority of orofacial pains that cannot be classified using current diagnostic criteria[5,6] do not correspond to an obvious physical or pathologic abnormality. Early diagnosis is therefore rare, as current medical practice is focused on identifying underlying abnormal pathology for reported symptoms. Patients are referred from clinician to clinician within dental and medical specialties in search of a pathologic cause. A diagnosis of idiopathic orofacial pain is often reached only after multiple tests and treatments have failed to improve the patient's condition, ie, after much wasted time and effort. The psychosocial burden of chronic orofacial pain has been well documented; studies in clinical settings show that subjects with chronic orofacial pain reported higher levels of psychosocial disability compared with those who reported acute orofacial pain.[7] Early identification could have a huge clinical and economic impact in the management of these conditions by allowing appropriate referral and management at the outset.

The authors have previously developed and validated a questionnaire-based tool[8] for classifying self-reported orofacial pain in population-based studies. However, this tool placed orofacial pain conditions likely to have an underlying pathology and those likely to be idiopathic in the same category. It was therefore not possible to use this tool without further modifications to identify subjects who reported idiopathic orofacial pain. In addition, the classification criteria were too broad and included headaches within the definition of orofacial pain.

The aim of the current study was therefore to develop and validate a questionnaire-based tool that would enable classification of idiopathic orofacial pain in the general population. Specific objectives were *(1)* to determine whether orofacial pain could be accurately classified as idiopathic, dentoalveolar, or musculoligamentous based on distinct characteristics reported on a self-administered questionnaire and *(2)* to estimate the prevalence of idiopathic orofacial pain in the general population.

Materials and Methods

Study Design and Participants

The study was a population-based cross-sectional survey of 4,200 randomly selected adults aged 18 to 75 years registered with a general medical practice in Handforth (borough of Macclesfield, northwest England). Because 95% of the adult population within the United Kingdom is registered with a general practitioner, and the selected general medical practice serves an area with mixed socio-demographic characteristics,[9] the sampling frame from the current study was representative of its source population. Subjects were requested to complete a postal questionnaire between September 2003 and June 2004. The study questionnaire can be accessed in appendix 9 of a published thesis.[10] Ethical approval of the survey was granted by the Macclesfield Research Ethics Committee, East Cheshire NHS Trust. The methodology and response rates for the population-based study have been described in detail previously.[11]

All subjects who reported facial pain (defined as pain in the face, mouth, or jaws which had been present for a day or longer in the past month) on the study questionnaire were invited to be interviewed and examined at their general medical practice, home, or other suitable location by a trained interviewer. Interoperator reliability using a structured history for the interview was established in a previous study.[8] Briefly, the interoperator reliability was established by comparing the diagnoses of 2 relatively inexperienced clinical interviewers (VA and RC) with those of facial pain experts (JMZ, PA, and ME). The structured history can be found as an appendix to a published thesis.[10] There was substantial agreement between the facial pain experts (kappa = 0.78) and between the clinical examiners and experts (kappa = 0.80).[8] The structured history was therefore deemed a valuable tool in diagnosing painful conditions of the face. The interviewer (VA) was blinded to the questionnaire responses of the interviewed subjects. All subjects were classified upon interview and clinical examination into 1 of 4 categories based on their chief orofacial pain complaint: musculoligamentous and soft tissue (MLST), neuralgic/vascular, dentoalveolar, and idiopathic. The orofacial pain conditions that were assigned to each category are shown in Table 1. Conditions that could not be assigned a diagnosis using a structured protocol (available upon request) that encompassed current diagnostic criteria for facial pain[5,6,12,13] were classified as idiopathic. These categories were developed a priori by a facial pain expert (JMZ) and are described in detail in appendix 5 of a published thesis.[10]

Table 1 Specific Diagnoses of Orofacial Pains Within Each Category

Diagnoses	n	%
Neuralgic/vascular	20	10
Neuropathic pain	3	15
Trigeminal neuralgia	3	15
Atypical trigeminal neuralgia	1	5
Glossopharyngeal neuralgia	1	5
Chronic tension headache	2	10
Migraine	10	50
Musculoligamentous/soft tissue (MLST)	44	23
Burning mouth syndrome	1	2
Temporomandibular joint pain	25	57
Recurrent oral ulceration	7	16
Lichen planus	2	5
Herpes labialis	1	2
Other rare conditions*	8	18
Dentoalveolar	57	30
Periodontal disease	9	16
Chronic pulpitis	20	35
Reversible pulpitis	2	4
Periapical periodontitis	2	4
Sinusitis	10	18
Dry socket	1	2
Dentine sensitivity	4	7
Pericoronitis	7	12
Sore dentures	1	2
Other rare condition (parotitis)	1	2
Idiopathic	72	37
Total	193	100

*Ear infection (n = 2), eye infection, lymphadenopathy, soft tissue abscess, sore throat (n = 2), and urticaria.

Study Questionnaire

The study questionnaire included validated scales that inquired about the reporting of other unexplained symptoms, psychologic factors, disability related to orofacial pain, and questions for classification of orofacial pain. These are described here, but further details can be found in appendix 9 of a published thesis[10] and a previously published article.[11]

Measurement of Other Frequently Unexplained Pain Syndromes

Chronic widespread pain was defined according to the American College of Rheumatology criteria for classification of widespread pain.[14] For the purpose of the current analysis, headache pain was excluded from the definition of chronic widespread pain, because orofacial pain was considered a separate outcome. Irritable bowel syndrome was measured according to the Rome II criteria,[15] and chronic fatigue was defined by fatigue scores of at least 8 on the Chalder fatigue scale[16] and the presence of such fatigue for 6 months or longer.

Measurement of Psychologic Factors

Psychologic distress was measured using the Hospital Anxiety and Depression (HAD) scale.[17] Scores for anxiety and depression were categorized into 3 groups: a score of 0 to 7 represented a noncase; a score of 8 to 10 indicated a borderline case, and a score of 11 to 21 represented a probable case of anxiety/depression.

Maladaptive response to illness was measured using the Health Anxiety Questionnaire (HAQ),[18] while sleep disturbance was measured using a validated 4-item sleep scale.[19] Because distributions of scale scores were not Gaussian, health anxiety and sleep scores were divided into tertiles to provide 3 equally sized groups for analysis.

Somatization was measured using a somatic symptoms checklist,[20] while a list of threatening experiences[21] was used to gather information on recent adverse life events. Because the distribution of somatic symptoms, scores (range, 0 to 5) and adverse life events, scores (range, 0 to 9) were highly skewed, subjects with scores of 0 were used as the reference category, while subjects with scores greater than 0 were dichotomized. Further,

for the somatic symptoms scale, the question relating to pain symptoms was excluded, because pain was considered a separate outcome.

Classification Questionnaire for Orofacial Pain

The classification questionnaire used for the current study was a modified version of a questionnaire developed in a previous population-based study.[8] The original questionnaire was modified, as the previous study identified a large proportion of headaches that were not related to the orofacial region. Further, the previous questionnaire was not designed to classify idiopathic orofacial pain; such pain was grouped with other pains in the orofacial region. The classification questionnaire can be found in appendix 2 of a published thesis.[10] The classification included the following aspects of orofacial pain:

Duration. Duration of orofacial pain was measured using 2 questions:

1. "During the past month have you had pain in your face, mouth or jaws which has lasted for 1 day or longer?" This question has been used successfully in identifying subjects with chronic widespread pain[22] and enables the elimination of trivial pain symptoms. Subjects are also more likely to recall symptoms over this time period more accurately than over longer time periods, such as 6 to 12 months. All subjects who answered "yes" to this question were eligible for interview.
2. "Did this pain begin more than 3 months ago?" This is in accordance with the International Association for the Study of Pain[5] definition of chronic pain (pain which persists for 3 months or longer).

For the purpose of analysis, pain duration was dichotomized (presence or absence of orofacial pain).

Site. Site of orofacial pain was measured using 3 blank mannequins of the face and an intraoral diagram including the teeth and associated soft tissues to determine the exact intraoral location of the pain.

The previous questionnaire[8] used 9 preshaded diagrams to determine pain distribution and asked patients to circle the picture that illustrated the location of their maximum pain. However, it was assumed that all pain conditions can be accurately represented and mapped with preshaded diagrams. Frequently, patients with chronic pain conditions have pain at multiple sites and in varying patterns. It may not be possible to use preshaded diagrams to represent the site of such pain if the pain follows a different pattern than that shown on the diagram. Indeed, this proved to be the case when the preshaded diagrams were used previously.[8] Some subjects had indicated that the preshaded mannequins did not represent the site of their orofacial pain. In addition, some of the preshaded diagrams specifically alluded to headache pain.

Therefore, to restrict self-report pain as far as possible to the orofacial region, unshaded extraoral mannequins of the head and neck were used. Subjects were asked to shade on the mannequins the site of their pain. Furthermore, an intraoral diagram including the teeth and associated soft tissues was added to determine the exact intraoral location of the pain. There are numerous structures in the oral cavity, and shading the mouth on a diagram of the face is insufficient evidence to indicate that a subject's pain has arisen from the teeth or oral soft tissues. In addition, chronic orofacial pain conditions such as burning mouth syndrome commonly include a painful, burning sensation in the oral soft tissues, and a diagram of the oral cavity would also help in identifying such cases.

The extraoral region of the face was then divided anatomically into 15 sites; the intraoral region was divided into a further 16 sites. The information provided by the facial mannequins was thus coded using this template of 31 sites. This method of identifying and coding pain has been successfully used previously to investigate the prevalence of regional and widespread pain syndromes.[22] The site of pain was analyzed according to the number of pain sites reported. The distribution of the number of sites shaded on the facial pain mannequins was not Gaussian. Rather, patients were placed in 1 of 3 categories: *(a)* 1 to 3 sites shaded, *(b)* 4 or 5 sites shaded, and *(c)* 6 or more sites shaded.

Intensity, Disability, and Consulting Behavior. For the purpose of analysis, pain intensity and disability were also dichotomized. Low levels of pain intensity encompassed scores of 0 to 5 on the visual analog scale (VAS), while high pain intensity ranged from 6 to 10. The corresponding disability scores were 0 to 3 (low levels) and 4 to 31 (high levels) on a recently validated orofacial pain disability scale.[7]

Information on consulting behavior of subjects with orofacial pain was analyzed according to whether subjects had consulted any health-care professionals and the number of health-care pro-

fessionals that they had consulted. Number of health-care professionals consulted was treated as a continuous variable (range, 1 to 5) in the univariate and multivariate analyses.

Pain Descriptors and Pattern. Pain descriptors for orofacial pain were measured using an adaptation of the short-form McGill Pain Questionnaire created by a facial pain expert (JMZ) for patients reporting orofacial pain.[23] Pain pattern was ascertained using a series of 17 yes/no questions on pain pattern and associated symptoms adapted (by JMZ) from a questionnaire designed by Hapak et al[24] to classify orofacial pain in a tertiary setting. Adaptations were made by removing repeated questions from the original questionnaire and by removing 2 other questions that were not useful in classifying orofacial pain: *(1)* "my principal pain is at its maximum at the beginning" and *(2)* "my principal pain makes me sick to my stomach (nauseated)." These statements are nonspecific and may be associated with a variety of facial pain conditions, making them redundant in discriminating between these conditions. Two other questions that did not specifically inquire about orofacial pain were also left out, and 3 questions that were all essentially asking about jaw joint pain were merged into 1 question: "My pain gets worse the more I move my jaw when eating, chewing and/or talking."

The instrument by Hapak et al[24] had sensitivities of 78.7%, 78.9%, and 37.5% for distinguishing the MLST, neuralgic, and dentoalveolar groups, respectively, and specificities of 81.5%, 78.2%, and 97% for the same 3 groups. These results indicate that the sensitivity of this questionnaire for the identification of dentoalveolar pain was low. Based on this, 4 specific questions were included to identify the commonly encountered dentoalveolar pains, namely toothache and sinusitis:

1. "My pain arises from my tooth/teeth."
2. "My pain gets worse when I bend my head forwards to my knees."
3. "My pain is associated with a running nose."
4. "My pain is associated with red/watery eyes."

Oral Health Factors

Questions were also included on other oral health factors, such as facial trauma and teeth grinding, which were analyzed as dichotomous (yes/no) variables. These were not used to classify orofacial pain and are not part of the classification questionnaire.

Statistical Analysis

Because the dependent variable (interview group) had 3 categories (MLST, dentoalveolar, and idiopathic) which had no natural ordering, multinomial logistic regression was used. Initially, χ^2 tests for statistical significance were used to determine whether there were differences in questionnaire variables between the 3 groups. A univariate analysis using multinomial logistic regression was also conducted to provide odds ratios (OR) with 95% confidence intervals (CIs) for each questionnaire variable, using the largest (dentoalveolar category) as the reference.

To exclude the role of chance and avoid omission of variables that were clinically important in distinguishing between the 3 categories, the significant variables ($P \leq .05$; χ^2 test) or those with OR in the univariate analysis for idiopathic and MLST pain of greater than 4.0 or less than 0.25 were entered into a multivariate model. However, 2 variables (tingling pain and pain located on skin) that are clinically important and were statistically significant had to be excluded, as the reference category contained no subjects reporting these characteristics, and this made the OR for the other categories infinite. Finally, age and gender were "forced" into the model. Although they were not statistically significant, their role as potential confounders could not be ignored.

Finally, a forward stepwise selection was used to construct the multivariate model. The criteria for selecting variables that were entered into the model have been discussed. Similarly, criteria used to decide which of these variables were retained in the model were based on a biologically plausible and mathematically stable model. Therefore, $P < .1$ was used as the significance level for retention of variables in the forward stepwise model.[25] Predictive probabilities were used to calculate the sensitivity and specificity of the final model at predicting membership into categories identified by clinical interviews. All analyses were carried out using STATA, version 8.[26]

Results

Interview Response Rates

Of those who responded (n = 2,505), 299 (12%) subjects reported oroxfacial pain (pain in the face, mouth, or jaws which had lasted for a day or longer) and were therefore eligible for interview. Of these, 224 (75%) consented to further contact

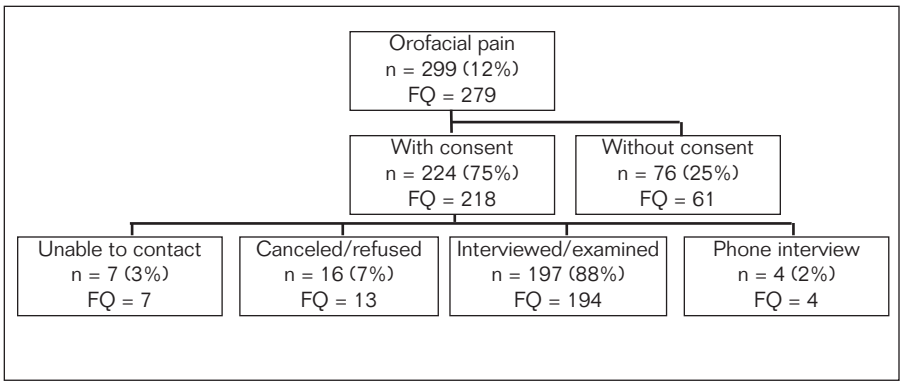

Fig 1 Interview and examination response rates. FQ = full questionnaire.

on the questionnaire and were invited for an interview and examination. However, 16 (7%) refused an interview, 7 (3%) could not be reached, and 4 (2%) were interviewed over the telephone. Therefore, 197 (88%) of those who agreed to be contacted were interviewed and examined (Fig 1); 194 (99%) of these had fully completed the study questionnaires, and this was the sample used for further analysis.

However, upon examination of the interview data, it was noted that 1 subject had been classified into 2 categories because of the presence of 2 distinct pains in the orofacial region. That subject was therefore excluded from the analysis. The sample was thus reduced to 193 subjects. In addition, the primary interest was in orofacial pain, which by definition is pain inferior to the orbitomeatal line, superior to the neck, and anterior to the ears. A small number of subjects were identified who reported headaches. According to the examination protocol these were placed in the neuralgic/vascular category (Table 1). Of the subjects in the neuralgic/vascular category (n = 20), 12 (60%) were identified by interview as having headache pain (10 migraine and 2 chronic tension headache). Therefore, to avoid spurious findings (the study was investigating pain in the mouth and face), subjects who reported headache were excluded from further analysis. This reduced the number of subjects categorized as neuralgic/vascular to 8. Because these numbers were too small to analyze meaningfully, and because the primary interest was in idiopathic pain, it was decided that the neuralgic/vascular category would be excluded from further analysis. Therefore, for the purpose of analysis, interviewed subjects (n = 173) were placed in 3 categories: MLST, dentoalveolar, and idiopathic.

Prevalence of Subtypes of Orofacial Pain Identified by Interview

Of those subjects who were interviewed (n = 173), 57 (33%) were classified as having dentoalveolar pain, 44 (25%) were classified as having MLST pain, and 72 (42%) were classified as having idiopathic pain. There were no significant gender differences between these categories; however, those in the idiopathic category had increased odds of being in the oldest age category compared to the dentoalveolar category (Table 2).

Orofacial Pain Subtypes and Reporting of Psychologic Factors

There were no remarkable differences in the distribution of psychologic factors between the 3 orofacial pain categories (Table 2). The median psychologic scale scores for each measure were roughly the same across orofacial pain subtypes. However, those who reported idiopathic pain had increased odds of reporting that their pain was worse when stressed and also had significantly (P = .002; Wilcoxon rank sum test) higher psychologic disability scores (median 2; interquartile range [IQR] 1–5) compared to those in the dentoalveolar group (median 1; IQR 0–2).

Pain Severity Within Orofacial Pain Subtypes

Subjects who were classified as having idiopathic pain reported higher scores for 2 of 3 pain-severity measures compared with those reporting dental or MLST pain. They had increased odds of reporting pain at multiple sites and higher disability (Table 3). There was no difference in pain intensity between those with idiopathic pain and those with dental pain (Table 3).

Table 2 Associations of Age, Gender, and Psychologic Factors with Orofacial Pain Subtypes

			Orofacial pain category*			
			Idiopathic		MLST	
Variable	*P*	Range	OR	95% CI	OR	95% CI
Age	.058	18–42	1	Ref	1	Ref
		43–55	1.1	0.5–2.6	0.7	0.3–1.7
		56–75	3.4	1.3–8.8	2.0	0.7–5.5
Gender	.751					
M			1	Ref	1	Ref
F			1.3	0.6–2.6	1.3	0.6–3.0
Health anxiety (HAQ)	.300	0–7	1	Ref	1	Ref
		8–13	0.9	0.3–2.6	0.5	0.2–1.6
		14–59	0.7	0.3–1.9	0.4	0.1–1.0
Depression (HAD)	.477	0–7	1	Ref	1	Ref
		8–10	0.4	0.1–1.1	0.3	0.1–1.1
		11–21	1.8	0.7–5.0	0.8	0.2–2.9
Anxiety (HAD)	.195	0–7	1	Ref	1	Ref
		8–10	0.9	0.3–2.3	0.3	0.1–0.9
		11–21	1.4	0.6–3.2	0.5	0.2–1.4
Sleep disturbance	.195	0–3	1	Ref	1	Ref
		4–7	1.2	0.4–3.5	3.0	0.9–10.4
		8–20	1.4	0.6–3.5	1.9	0.6–5.8
Somatic symptoms	.716	0	1	Ref	1	Ref
		1	1.9	0.7–4.8	1.4	0.5–4.0
		2–6	1.7	0.7–4.3	1.9	0.7–5.2
Adverse life events	.760	0	1	Ref	1	Ref
		1	1.7	0.6–4.4	1.0	0.3–3.1
		2–9	1.3	0.6–3.0	1.6	0.6–3.9

*Dentoalveolar was the reference category.

Table 3 Associations Between Consultations and Pain Severity with Orofacial Pain Subtypes

			Orofacial pain category*			
			Idiopathic		MLST	
Variable	*P*	Range	OR	95% CI	OR	95% CI
No. of pain sites	.050	1–3	1	Ref	1	Ref
		4–5	2.1	0.8–5.1	0.8	0.3–2.0
		≥ 6	2.5	1.0–6.0	0.6	0.2–1.6
Pain Intensity (VAS)	.235	0–5	1	Ref	1	Ref
		6–10	1.1	0.5–2.4	0.5	0.2–1.3
Disability	.018	0–3	1	Ref	1	Ref
		3–31	3.4	1.5–7.3	1.5	0.6–3.5
Consulting behavior	.323	No	1	Ref	1	Ref
		Yes	0.5	0.2–1.1	0.7	0.3–1.7
No. of HCW visited	.006	1–5	5.2	2.1–13.0	3.0	1.2–7.8

*Dentoalveolar was the reference category. HCW = health-care workers.

Consultation Behavior Among Orofacial Pain Subtypes

Subjects with idiopathic orofacial pain had decreased odds of consulting a health-care professional for their pain. However, when they did consult, these subjects had significantly increased odds of seeking advice from multiple health-care workers for their pain (Table 3).

Orofacial Pain Subtypes and Reporting of Other Oral Health Factors

Subjects with idiopathic pain had increased odds of being registered with a dentist compared to those with dentoalveolar pain (Table 4). They also had increased odds of reporting grinding of their teeth and were more likely to have had their teeth grinding verified by their dentist or a family member.

Table 4 Associations Between Symptom Reporting and Oral Health with Orofacial Pain Subtypes

		Orofacial pain category*			
		Idiopathic		MLST	
Variable	*P*	OR	95% CI	OR	95% CI
Chronic OFP	.007	3.2	1.4–7.2	1.1	0.5–2.6
CWP	.011	3.2	1.4–7.4	2.1	0.8–5.4
IBS	.023	2.2	0.9–5.2	0.7	0.2–2.1
Chronic fatigue	.496	1.3	0.6–2.9	0.8	0.3–2.1
No. of symptoms	.014	1.7	1.1–2.5	1.1	0.7–1.7
Dental registration	.183	2.6	0.8–8.5	5.7	1.1–29.9
Check-ups	.099	1.0	0.4–2.4	3.5	1.0–12.0
Tooth grinding	.352	2.0	0.9–4.5	1.6	0.7–4.1
Verification of tooth grinding	.125	2.8	1.2–6.7	1.7	0.6–4.6
Teeth fit	.748	0.9	0.4–1.9	0.8	0.3–1.8
Teeth missing	.390	0.9	0.4–1.9	1.6	0.6–3.9
Trauma	.025	2.6	1.1–5.8	1.0	0.4–2.7

*Dentoalveolar was the reference category. CWP = chronic widespread pain; IBS = irritable bowel syndrome; OFP = orofacial pain.

Table 5 Associations of Pain Descriptors to Orofacial Pain Subtypes (short form McGill)

		Orofacial pain category*			
		Idiopathic		MLST	
Variables	*P*	OR	95% CI	OR	95% CI
Throbbing	< .001	0.3	0.1–0.7	0.3	0.1–0.7
Shooting	.112	0.6	0.2–1.5	0.3	0.1–0.9
Stabbing	.180	4.5	1.1–17.9	2.0	0.4–9.8
Sharp	.668	0.6	0.2–1.6	0.7	0.3–2.0
Gnawing	.531	0.5	0.2–1.3	0.8	0.3–2.3
Burning	.091	4.5	0.9–21.4	4.0	0.8–21.1
Tingling	.004	∞		∞	
Aching	.005	3.8	1.8–8.2	1.7	0.7–3.9
Tender	.576	1.0	0.5–2.2	1.4	0.6–3.2
Tiring	.787	1.6	0.6–4.7	1.1	0.3–3.7
Frightful	.150	4.1	0.5–38.8	1.0	0.1–17.0
Vicious	.744	2.3	0.6–8.7	1.5	0.3–6.6
Miserable	.094	3.6	1.2–10.9	2.4	0.7–8.2
Unbearable	.107	1.2	0.4–3.1	0.2	0.1–1.1
Nagging	< .001	3.1	1.4–7.0	0.5	0.2–1.4

*Dentoalveolar was the reference category.

Those with idiopathic pain also had increased odds of reporting facial trauma.

Orofacial Pain Subtypes and Reporting of Other Pain Symptoms

Subjects in the idiopathic category had increased odds of reporting chronic orofacial pain, chronic widespread pain, and irritable bowel syndrome compared to the dentoalveolar category (Table 4). Further, those in the idiopathic category also had increased odds of reporting multiple symptoms (Table 4).

Pain Descriptors Among Orofacial Pain Subtypes

The most frequently reported descriptors were aching (50%), tender (36%), and throbbing (35%). Some descriptors were rarely reported, for example, frightful (5%), tingling (9%), and stabbing (10%). Subjects with idiopathic pain had increased odds of reporting stabbing, burning, aching, frightful, miserable, nagging, and tingling pains compared with those in the dentoalveolar category. The OR for tingling pain is indicated as infinite in Table 5, as there were no subjects who reported this characteristic for dentoalveolar pain. Further, those with idiopathic pain also had decreased odds of reporting throb-

Table 6 Associations of Pain Pattern Among Orofacial Pain Subtypes

		Orofacial pain category*			
		Idiopathic		MLST	
Variables	*P*	OR	95% CI	OR	95% CI
Pain intensity					
Constant	.602	1.1	0.3–3.8	0.5	0.1–2.8
Intermittent	.719	1.3	0.6–2.8	1.1	0.5–2.5
Clusters	.255	1.5	0.6–3.8	0.5	0.2–1.9
Worse with time	.987	1.0	0.3–3.5	1.0	0.3–4.1
Pain location					
On skin	.072	∞	–	∞	–
From teeth	< .001	0.1	0.04–0.2	0.1	0.02–0.2
Unilateral	.022	2.3	1.1–4.9	0.9	0.4–2.2
Either side	.356	1.8	0.6–4.9	2.4	0.8–7.1
Bilateral	.006	9.7	2.1–45.2	5.1	0.9–27.0
Associated symptoms					
Begins on light touch	.876	0.9	0.1–6.8	1.4	0.2–10.3
Begins with hot/cold	.004	0.1	0.04–0.5	0.4	0.1–1.1
Worse with stress	< .001	15.7	4.2–57.9	4.3	1.0–17.65
Worse on jaw movement	.005	0.4	0.2–1.0	2.0	0.8–4.6
Worse bending forward	.035	1.8	0.6–5.4	0.2	0.02–1.7
Better heat/cold	.004	4.2	0.9–20.3	0	0
Running nose	.066	0.3	0.1–1.7	0	0
Red eyes	.956	0.8	0.2–4.3	0.9	0.1–5.5

*Dentoalveolar was the reference category.

bing, shooting, sharp, or gnawing pain, compared with the dentoalveolar category (Table 5).

Pain Pattern Among Orofacial Pain Subtypes

Subjects with idiopathic pain had increased odds of reporting pain that was unilateral and bilateral compared to those with dentoalveolar pain (Table 6). They also had increased odds of reporting pain that worsened with stress and pain that improved when heat or cold was applied to the face. As expected, those with idiopathic pain had decreased odds of reporting pain that arose from their teeth, pain that started on eating hot/cold foods, and pain that was worse on jaw movement compared to those with dentoalveolar pain (Table 6). Also, as expected, those with MLST pain had increased odds of reporting pain that got worse on jaw movement when compared to those with dentoalveolar or idiopathic pain (Table 6).

Final Multivariate Model

Overall, 22 variables were selected from the univariate analysis for the forward stepwise procedure. The final model thus generated (Table 7) contained 10 variables, including age and gender (the 2 variables "forced" into the model). Based on this model, compared to subjects classified as having dentoalveolar pain, those with idiopathic pain had increased odds of being older and female. They also had increased odds of reporting aching and nagging pain, which was worse when they were stressed. Finally they had increased odds of reporting unilateral pain and pain at > 6 sites (Table 7). As expected, those with idiopathic pain had reduced odds of reporting throbbing pain, pain that arose from their teeth and pain that was worse on jaw movement.

Sensitivity and Specificity of the Final Model

The highest sensitivity was for the idiopathic group; that is, the model was able to correctly classify 76% of those with idiopathic pain (Table 8). The sensitivity for the dentoalveolar category was 75%, while that of the MLST category was 62% (Table 8). The corresponding specificities were 78% for the idiopathic category, 87% for the dentoalveolar category, and 88% for the MLST category. Overall, there was very good agreement between the classification predicted by the model and that identified by clinical examination (kappa = 0.6). Based on probabilities equal to the overall proportions shown in Table 8, one would expect the model to agree randomly on 34% of the subjects. In fact, the model agreed on 72% of subjects or 60% (kappa = 0.6) of the way between random and perfect agreement. The hypothesis that there is random agreement between the model and clinical classification can therefore be rejected.

Table 7 Final Multivariate Model

	Orofacial pain category*			
	Idiopathic		MLST	
Variables	OR	95% CI	OR	95% CI
Age				
18–42	1	Ref	1	Ref
43–55	2.3	0.6-8.7	0.6	0.2-2.3
56–75	5.7	1.1-29.6	1.1	0.2-5.3
Gender				
M	1	Ref	1	Ref
F	2.3	0.7-7.5	1.0	0.3-3.3
Pain from teeth				
No	1	Ref	1	Ref
Yes	0.05	0.0–0.2	0.01	0.004–0.1
Worse when stressed				
No	1	Ref	1	Ref
Yes	29.8	4.7–189.6	8.4	1.2–60.2
Worse on jaw movement				
No	1	Ref	1	Ref
Yes	0.4	0.1–1.7	7.3	2.0–27.1
Unilateral				
No	1	Ref	1	Ref
Yes	12.6	3.1–50.8	1.6	0.4–6.1
Nagging				
No	1	Ref	1	Ref
Yes	11.8	2.9–47.9	0.7	0.2–3.6
Aching				
No	1	Ref	1	Ref
Yes	4.9	1.5–15.8	1.9	0.6–6.0
Throbbing				
No	1	Ref	1	Ref
Yes	0.2	0.1–0.8	0.6	0.2–2.1
No. of pain sites				
1–3	1	Ref	1	Ref
4–5	0.9	0.2–3.8	0.3	0.1–1.1
> 6	2.4	0.5–11.1	0.1	0.03–0.7

*Dentoalveolar was the reference category.

Table 8 Comparison of Model-Based Classification to that Identified by Examination

	Classification by clinical examination		
Predicted model classification	Dentoalveolar 57 (33)	MLST 44 (25)	Idiopathic 72 (42)
Dentoalveolar 56 (32)	43 (75)	5 (11)	8 (11)
MLST 39 (23)	4 (7)	27 (62)	8 (11)
Idiopathic 74 (43)	8 (14)	11 (25)	55 (76)
Missing 4 (2)	2 (4)	1 (2)	1 (1)

Values in parentheses indicate percentages.

Prevalence of Idiopathic Orofacial Pain

Overall, 279 subjects with orofacial pain completed full questionnaires (Fig 1) and could therefore be classified into 1 of the 3 categories based on their questionnaire responses. One hundred ninety-four subjects were classified by clinical interview (Fig 1). This left 85 subjects who had completed full questionnaires but had not been interviewed. Based on the final model, 80 (94%) were classified by the model. Data were missing for 5 cases, and the model was therefore unable to classify these. The noninterviewed

subjects (n = 80) were therefore classified as follows: 31 (39%) in the dentoalveolar group, 27 (34%) in the idiopathic group, and 22 (27%) in the MLST group. Therefore, upon combining this predicted classification and that determined by interview, the prevalences of the types of orofacial pain in the present study were 4.3% for idiopathic pain, 3.8% for dentoalveolar pain, and 2.9% for MLST pain.

Discussion

This study has shown, for the first time, that idiopathic orofacial pain is frequently reported by the general population (prevalence 4.3%) and may be a distinct separate entity from other orofacial pain conditions that have an underlying pathologic cause. Further, the final multivariate model demonstrated questionnaire-based criteria that may provide a useful tool to screen for idiopathic orofacial pain in population-based studies, where clinical examination of subjects is not feasible.

The prevalence of orofacial pain in the current cross-sectional study (12%) was comparable to 2 other studies.[27,28] These studies were population-based self-report studies of facial pain conducted in the United States. Although 1 of the studies[29] did not encompass a socio-demographically mixed sample, the other[28] involved a national general health survey and encompassed a representative sample. The current study was conducted in the United Kingdom in a socio-demographically mixed population that was representative of its source population. The disease definition used in these studies was consistent with the current cross-sectional study; therefore, the results can be compared with confidence. Other studies have included trivial symptoms of toothache[29] or have included headaches in their definition.[30] Therefore, they are likely to have overestimated the prevalence of orofacial pain.

However, there are methodologic issues that warrant consideration. First, the present analysis is likely to have underestimated the prevalence of idiopathic orofacial pain. Conditions such as TMD often have no underlying pathology. These conditions were classified in the MLST category. However, there was only 1 case of burning mouth syndrome, and TMD was identified if subjects had associated symptoms that could be attributed to this diagnosis using current diagnostic criteria.[13] An underestimate of the prevalence by not including these conditions in the idiopathic category would only lead to a dilution of the strength of the associations observed in the analysis.

Second, there were null values for some pain characteristics in certain categories, such as tingling in the dentoalveolar category. Such variables were omitted in the final multivariate model to make it mathematically stable. One cannot discount the importance of such variables in distinguishing between orofacial pain subtypes, and although the final multivariate model had a high sensitivity for predicting subjects with idiopathic pain, there are likely to be other variables that can add to its discriminative power.

Third, although the study achieved a high participation rate, nonresponse could still have affected the results. Noninterviewed subjects (n = 85) were more likely to be younger males and to report pain that arose from their teeth and throbbing pain—all characteristics of dental pain. Indeed the model classified most of the noninterviewed subjects into the dentoalveolar category, while the remainder were classified among the idiopathic and MLST categories, as the variables that define these pain subtypes were less frequently reported by the noninterviewed subjects. Therefore, although the noninterviewed subjects had different demographic and pain characteristics compared to those who were interviewed, this was taken into account by the model during classification. These differences did not affect the classification of nonparticipants. A detailed description of noninterviewed subjects is presented in Table 9.14 of a published thesis.[10]

The present study had several strengths. Current diagnostic criteria for idiopathic orofacial pain tend to rely on clinic-based studies and expert opinion to classify such pain.[6,12,31] Clinical studies represent only the most severe and intractable cases, those selectively referred from primary care, and are therefore unlikely to represent the full spectrum of idiopathic orofacial pain. The current study was population based. It is therefore likely to have encompassed both mild and severe cases of idiopathic pain. Furthermore, the current study used patient responses to classify orofacial pain subtypes. This is more accurate than expert opinion, as it is the patient who is experiencing the pain and who can thereby relate precisely to its description and associated features, although expert-designed criteria were used as the gold standard to validate the classification instrument. An additional strength of the study is that the results are unlikely to have been affected by misclassification bias because the interviewer was blinded to the patient responses before clinical examination. In clinical studies, the clinician is aware of the patient's exposure status prior to the examination.

Finally, the current study only included those pains that were confined to the orofacial region. Headache pain was excluded from the analysis, as the primary interest was in pain from the orofacial region. Previous studies that have investigated orofacial pain in its entirety often include headaches, which are in close proximity to the orofacial region and are often difficult to differentiate from orofacial pain. Such studies are notable for the high prevalence rates obtained for orofacial pain.[29,30]

The results from the current classification are comparable to those of previous studies.[1,6,12,32–39] The present study found a number of similarities to current diagnostic criteria for idiopathic orofacial pain.[6,12] Subjects with idiopathic orofacial pain were more likely to report aching and nagging pain and less likely to report throbbing pain, which is a common descriptor for toothache. In agreement with the International Headache Society (IHS) criteria,[6,12] those with idiopathic pain were more likely to report pain in multiple sites. In addition, the finding that subjects with idiopathic orofacial pain reported pain that was both unilateral and bilateral may indicate that this pain condition is poorly localized—a finding supported by the IHS criteria.

Not surprisingly, idiopathic pains were less likely to arise from the teeth or begin on eating or drinking hot or cold foods, as these patterns indicate characteristics of caries-induced dental pain. In addition, these pains were less likely to get worse during jaw movement—a characteristic of TMD that was classified in the MLST category.

Further, subjects with idiopathic pain were more likely to report pain that was chronic, that is, present for 3 months or longer. This is also in agreement with the IHS criteria,[6,12] which establish that idiopathic facial pain is persistent.

Female predominance of subjects with idiopathic orofacial pain, as evidenced in the present study, is in agreement with clinically based studies that have investigated characteristics of atypical facial pain.[36–39] Such studies also support the present finding that subjects with idiopathic orofacial pain are more likely to report other pain conditions, such as chronic widespread pain and irritable bowel syndrome, and multiple symptoms compared to those who had orofacial pain with an identifiable pathology. This may support the hypothesis that idiopathic orofacial pain may be part of a wider spectrum of unexplained disorders.

Subjects with idiopathic orofacial pain were more likely to indicate that their pain worsened when they were stressed. They also had higher psychologic disability scores. Clinical studies have implicated that psychologic factors are a part of idiopathic orofacial pain conditions such as atypical facial pain.[32,38] The International Association for the Study of Pain[5] also classifies atypical facial pain as pain of psychogenic origin. Aspects of psychologic distress have also been shown to be important in the persistence of orofacial pain in its entirety.[35]

Subjects with idiopathic orofacial pain were more likely to report having experienced facial trauma, which is in agreement with the IHS criteria and studies in clinical settings,[34] which have implicated the role of trauma in persistent orofacial pain. Not surprisingly, those with idiopathic pain were more likely to have consulted a larger number of health-care professionals for their pain. This finding is supported by several studies[1,32,33] and is a characteristic of subjects presenting with unexplained pain conditions to any medical specialty.

Using these characteristics, the present analysis has developed a questionnaire-based classification of idiopathic orofacial pain that can be used as a screening tool in population-based studies of this condition. The instrument had a high sensitivity (76%) and specificity (78%) for classifying those with idiopathic orofacial pain, and there was substantial agreement (kappa = 0.6) between classification predicted by the instrument and that observed by the gold standard of clinical examination. Further work is needed to establish the validity of this instrument and classification model in an independent population.

This screening tool should allow future prospective studies to classify idiopathic orofacial pain and determine etiologic factors leading to its onset. That is, it can be used to determine whether potential etiological factors measured at baseline in subjects who were free of idiopathic orofacial pain led to the onset of this condition at follow-up. This will pave the way for intervention studies on whether intervention early in the natural history of idiopathic orofacial pain can lead to improved outcomes.

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References

1. Madland G, Feinmann C. Chronic facial pain: A multidisciplinary problem. J Neurol Neurosurg Psychiatry 2001; 71:716–719.
2. Wessely S, Nimnuan C, Sharpe M. Functional somatic syndromes: One or many? Lancet 1999;354(9182):936–939.
3. Harness DM, Donlon WC, Eversole LR. Comparison of clinical characteristics in myogenic, TMJ internal derangement and atypical facial pain patients. Clin J Pain 1990; 6(1):4–17.
4. Woda A, Tubert-Jeannin S, Bouhassira D, et al. Towards a new taxonomy of idiopathic orofacial pain. Pain 2005; 116:396–406.
5. Merskey H, Bogduk N, International Association for the Study of Pain. Classification of Chronic Pain: Descriptions of Chronic Pain Syndromes and Definitions of Pain Terms, ed 2. Seattle: IASP Press, 1994.
6. Classification and diagnostic criteria for headache disorders, cranial neuralgias and facial pain. Headache Classification Committee of the International Headache Society. Cephalalgia 1988;8(suppl 7):1–96.
7. Aggarwal VR, Lunt M, Zakrzewska JM, Macfarlane GJ, Macfarlane TV. Development and validation of the Manchester orofacial pain disability scale. Community Dent Oral Epidemiol 2005;33:141–149.
8. Macfarlane TV, Blinkhorn AS, Craven R, et al. Can one predict the likely specific orofacial pain syndrome from a self-completed questionnaire? Pain 2004;111:270–277.
9. Macclesfield District Profile: Electoral Wards in Macclesfield. Available at http://www.cheshire.gov.uk/NR/rdonlyres/86389E3D-321E-4FB5-B1A8-AD16F3F4737F/0/MacclesfieldDistrict.pdf
10. Aggarwal VRK. Epidemiology of Chronic Orofacial Pain [thesis]. Manchester, UK: University of Manchester, 2006.
11. Aggarwal VR, McBeth J, Zakrzewska JM, Lunt M, Macfarlane GJ. The epidemiology of chronic syndromes that are frequently unexplained: Do they have common associated factors? Int J Epidemiol 2006;35:468–476.
12. International Classification of Headache disorders, ed 2. Cephalalgia 2004 May 1;24(s1):23-136.
13. Dworkin SF, LeResche L. Research Diagnostic Criteria for Temporomandibular Disorders: Review, criteria, examinations and specifications, critique. J Craniomandib Disord 1992;6:301–355.
14. Wolfe F, Smythe HA, Yunus MB, et al. The American College of Rheumatology 1990 Criteria for the Classification of Fibromyalgia. Report of the Multicenter Criteria Committee. Arthritis Rheum 1990;33:160–172.
15. Thompson WG, Longstreth GF, Drossman DA, et al. Functional bowel disorders and functional abdominal pain. Gut 1999;45(suppl 2):1143–1147.
16. Chalder T, Berelowitz G, Pawlikowska T, et al. Development of a fatigue scale. J Psychosom Res 1993; 37:147–153.
17. Zigmond AS, Snaith RP. The hospital anxiety and depression scale. Acta Psychiatr Scand 1983;67:361–370.
18. Lucock MP, Morley S. The Health Anxiety Questionnaire. Brit J Health Psychol 1996;1(Part 2):137–150.
19. Jenkins CD, Stanton BA, Niemcryk SJ, Rose RM. A scale for the estimation of sleep problems in clinical research. J Clin Epidemiol 1988;41:313–321.
20. Othmer E, DeSouza C. A screening test for somatization disorder (hysteria). Am J Psychiatry 1985;142:1146–1149.
21. Tennant C, Andrews G. A scale to measure the stress of life events. Aust N Z J Psychiatry 1976;10:27–32.
22. Hunt IM, Silman AJ, Benjamin S, McBeth J, Macfarlane GJ. The prevalence and associated features of chronic widespread pain in the community using the "Manchester" definition of chronic widespread pain. Rheumatology (Oxford) 1999;38:275–279.
23. Melzack R. The short-form McGill Pain Questionnaire. Pain 1987;30:191–197.
24. Hapak L, Gordon A, Locker D, Shandling M, Mock D, Tenenbaum HC. Differentiation between musculoligamentous, dentoalveolar, and neurologically based craniofacial pain with a diagnostic questionnaire. J Orofac Pain 1994; 8:357–368.
25. Hosmer DW, Lemeshow S (eds). Applied Logistic Regression, ed 2. New York: John Wiley & Sons, 2000.
26. Stata [computer program]. Version Release 8.0. College Station, TX: Stata Corporation, 2003.
27. Von Korff M, Dworkin SF, LeResche L, Kruger A. An epidemiologic comparison of pain complaints. Pain 1988; 32:173–183.
28. Lipton JA, Ship JA, Larach-Robinson D. Estimated prevalence and distribution of reported orofacial pain in the United States. J Am Dent Assoc 1993;124(10):115–121.
29. Riley JL III, Gilbert GH. Orofacial pain symptoms: An interaction between age and sex. Pain 2001;90:245–256.
30. Macfarlane TV, Blinkhorn AS, Davies RM, Kincey J, Worthington HV. Orofacial pain in the community: prevalence and associated impact. Community Dent Oral Epidemiol 2002;30:52–60.
31. Elrasheed AA, Worthington HV, Ariyaratnam S, Duxbury AJ. Opinions of UK specialists about terminology, diagnosis, and treatment of atypical facial pain: A survey. Br J Oral Maxillofac Surg 2004;42:566–571.
32. Feinmann C, Harris M, Cawley R. Psychogenic facial pain: Presentation and treatment. Br Med J (Clin Res Ed) 1984;11;288:436–438.
33. Pfaffenrath V, Rath M, Pollmann W, Keeser W. Atypical facial pain—Application of the IHS criteria in a clinical sample. Cephalalgia 1993;13(suppl 12):84–88.
34. Benoliel R, Eliav E, Elishoov H, Sharav Y. Diagnosis and treatment of persistent pain after trauma to the head and neck. J Oral Maxillofac Surg 1994;52:1138–1147.
35. Macfarlane TV, Blinkhorn AS, Davies RM, Kincey J, Worthington HV. Predictors of outcome for orofacial pain in the general population: A four-year follow-up study. J Dent Res 2004;83:712–717.
36. Smith DP, Pilling LF, Pearson JS, Rushton JG, Goldstein NP, Gibilisco JA. A psychiatric study of atypical facial pain. Can Med Assoc J 1969;100:286–291.
37. Madland G, Newton-John T, Feinmann C. Chronic idiopathic orofacial pain: I. What is the evidence base? Br Dent J 2001;191:22–24.
38. Feinmann C, Harris M. The diagnosis and management of psychogenic facial pain disorders. Clin Otolaryngol 1984; 9:199–201.
39. Feinmann C, Harris M. Psychogenic facial pain. Part 1: The clinical presentation. Br Dent J 1984;156:165–168.