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ORIGINAL ARTICLE

The association between myofascial orofacial pain with and without referral and widespread pain

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ABSTRACT

Objectives: Pain referral on palpation has been suggested to be a clinical sign of central sensitization potentially associated with widespread pain conditions. Our aim was to evaluate if myofascial pain with referral is a better predictor for widespread pain when compared to no pain or local myofascial pain.

Materials and methods: Individuals at the Public Dental services in Västerbotten, Sweden, were randomly invited based on their answers to three screening questions for temporomandibular disorders (TMD). In total, 300 individuals (202 women, 20–69 yrs) were recruited, and examined according to the Diagnostic Criteria for TMD (DC/TMD) after completion of a body pain drawing. Widespread pain was considered present when seven or more pain sites were reported on the widespread pain index. A binary logistic regression model, adjusted for the effect of age and gender were used to evaluate the association between myofascial orofacial pain and widespread pain.

Results: Widespread pain was reported by 31.3% of the study sample. There was a 57.3% overlap with myofascial pain. Widespread pain was associated to myofascial orofacial pain with and myofascial orofacial pain (OR 4.83 95% CI 2.62–9.05 and OR 11.62 95% CI 5.18–27.88, respectively).

Conclusion: These findings reinforce the existing knowledge on the overlap between painful TMD and other chronic pain conditions.

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General practice dentistry; temporomandibular disorders; myofascial pain; widespread pain

Introduction

Chronic pain, that is, pain lasting beyond tissue healing time and ongoing for more than three months, is the most common reason for an individual to seek healthcare [1] and is associated with substantial societal costs [2] as well as suffering on the individual level [3]. Given that the pathophysiology behind chronic pain conditions is considered to differ from that of acute pain, chronic pain is regarded as a disease in itself rather than merely a symptom of tissue damage [4]. Therefore, individuals with chronic pain often report comorbid and complex psychosocial symptoms [5] that, in addition to the physical complaints, need to be addressed by the clinician. Thus, the management of patients with chronic pain is a challenge in health care.

The most common cause of chronic pain in the orofacial region is pain associated with temporomandibular disorders (TMD). This is an umbrella term encompassing musculoskeletal pain and dysfunction in the masticatory muscles and temporomandibular joint as well as the surrounding structures [6]. Painful TMD is defined as pain modified by jaw function [7,8], and is often associated with a dull ache of moderate pain intensity. The prevalence of chronic painful TMD is about 10% in the adult population and is most frequent among women [9]. For painful TMD, widespread pain is considered a risk factor not only for the onset but also for the maintenance of pain [10], and about one out of five individuals with TMD report concurrent widespread pain [11,12]. Widespread pain is characterized by pain in multiple body regions and has a prevalence of 10–15% in the general population [13]. In addition, it is associated with a variety of aggravating comorbidities, not only psychosocial disturbances but also physical symptoms and conditions such as cardiovascular disease [14] and even an increased risk of mortality [15]. Among individuals with widespread pain, approximately 75% report pain also in the orofacial region [16,17]. Patients with widespread pain and painful TMD share comorbidities, such as perceived stress [18], somatic symptoms [19], sleep disturbances [20], and depression [21], in addition to their chronic pain condition. Taken together, a multifaceted overlap between painful TMD and widespread pain is firmly established. This is also reflected in the term

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Chronic Overlapping Pain Condition (COPC) [22], and where TMD complaints have been suggested to be one of many chronic pain conditions without clarified pathophysiology [23,24]. However, in the clinical situation, comorbidity between orofacial pain and widespread bodily pain may be overlooked and diagnosed as a local TMD condition. Therefore, the relation and overlap between these conditions is of importance for the interpretation, diagnosis and management of orofacial pain. Taken together, among individuals with myofascial orofacial pain, it is important to be able to identify easily and correctly those with concurrent widespread pain [25].

Since 2014, the Diagnostic Criteria for TMD (DC/TMD) has provided a reliable and valid method for diagnosis of the most common TMDs. In the DC/TMD, the diagnosis of 'myalgia' can be further specified as the subdiagnosis 'myofascial pain with referral' in cases where pain spreads beyond the muscle border during palpation [7]. Such referred pain has been suggested related to a decrease in thresholds to nociceptive stimuli, potentially caused by general hyperalgesia and central sensitisation [26–28]. In dentistry, a diagnosis of referred pain was suggested to be relevant for differential diagnosis for the identification of pain in other anatomical locations such as muscular pain referred to the teeth [7]. However, there is a gap of knowledge regarding the association between a clinical diagnosis of pain referral and the presence of widespread pain in community samples with TMD.

Therefore, the aims were (i) to calculate the weighted prevalence of widespread pain using the Widespread Pain Index (WPI), and (ii) to evaluate if pain referral is a better predictor for widespread pain when compared to no pain or local myofascial in a community sample.

Material and methods

Study setting and participants

This case–control study was undertaken in Västerbotten, northern Sweden. In Sweden, about 80% of the adult population see their dentist on a regular basis and a majority do so at the Public Dental Health (PDH) services. In connection with a routine dental check-up, individuals answered three screening questions for TMD (3Q/TMD) [29]. The inclusion procedure has been described in detail previously [30]. In summary, this community sample was recruited from 7,831 individuals who attended the PDH services in Västerbotten during 2014 (Figure 1). Individuals with at least one affirmative answer to the 3Q/TMD were randomly recruited as individuals with a possible TMD, that is, 3Q-positives. In total, 152 3Q-positives and 148 3Q-negatives, that is, age and gender matched controls, were included [30]. The study was approved by the regional ethical board at Umeå university (ref no 2012-331-31 M). All participants signed an informed, written consent ahead of inclusion in the study. The STROBE statement with guidelines for reporting observational studies was followed [31].

Procedure

On the day of the examination, all individuals completed the DC/TMD symptom questionnaire, together with a full body pain drawing where they marked all areas of pain. Subsequently, all individuals were clinically examined by an examiner (AL), formally trained and calibrated according to the DC/TMD [7] (Kappa values; myofascial pain without referral 0.82 and myofascial pain with referral 0.88, respectively) and the individuals were diagnosed according to these

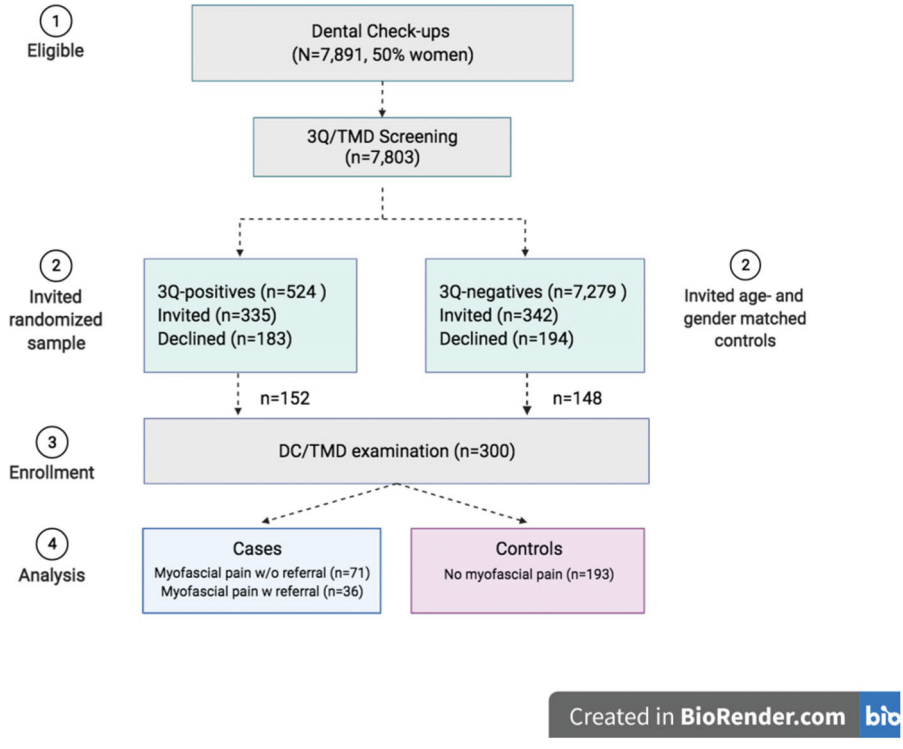

Figure 1. Flow-chart of the recruitment process.

criteria. The examiner was blinded to the group allocation but not to the study protocol.

In accordance with the DC/TMD and following the International Classification of Orofacial Pain, ICOP [25], myofascial orofacial pain was considered present when the following criteria were fulfilled: self-reported pain within the last 30 days, pain modified by function, and pain confirmed to the masseter or temporal muscles by the examiner together with familiar pain confirmed during jaw movement or muscle palpation during the clinical examination. In a second step, pain referral was considered present in the case of concurrent referred pain outside the muscle border during a 5-second muscle palpation. Based upon these criteria, all individuals were categorized into three groups; no myofascial pain (controls), myofascial pain without referral or myofascial pain with referral, respectively.

The number of painful sites were assessed from a whole-body pain drawing (0–19 pain sites, see Table 2 for details). The extremities and head/face were divided by left and right sides; for the neck, abdomen and upper- and lower back regions no division between left and right was made. In accordance with the widespread pain index [32,33], a cut-off at seven or more pain sites in the pain drawing was used.

Statistical analysis

Descriptive statistics were used to characterise the study sample, and non-parametric statistics with Chi^2 test and Mann–Whitney U-test were used for distributions and comparisons between groups. To account for the actual proportion of cases and controls in the general population, the population-based, weighted prevalence of widespread pain was calculated based on a normative prevalence of 4.8% 3Q positives among 20–69-year-olds in 2014. A binary logistic regression model, adjusted for the effect of age and gender, were used to evaluate the association to widespread pain (dependent variable) and the following categorical variables:

- no myofascial orofacial pain (reference)
- myofascial pain without referral
- myofascial pain with referral

A *post hoc* test using estimated marginal means to detect differences between the categories was applied. The statistical analyses were conducted in SPSS Statistics for Windows, Version 24.0. Armonk, NY: IBM Corp and in R (R v4.0.3, R Core Team). A *p*-value of <0.05 was regarded as statistically significant.

Results

In total, 202 women and 98 men, 20 to 69 years (mean age 38.7, SD 13.8), were included. In the total study sample ($n = 300$), 94 individuals (31.3%) reported widespread pain (Table 1). Widespread pain was significantly more frequent in women (37.6%) compared to men (18.4%) (Chi^2 test = 11.372, $p = .001$). There was no significant difference in age among individuals with and without widespread pain (Mann–Whitney U = 8,938.5, $p = .286$).

Table 1. Frequencies in the study sample ($n = 300$).

	Widespread pain ($n = 94$) n (%)	No widespread pain ($n = 206$) n (%)	Total ($n = 300$) n (%)
Gender			
Women (mean age 38.7, SD 13.3)	76 (25)	126 (42)	202 (67)
Men (mean age 38.7, SD 14.7)	18 (6)	80 (27)	98 (33)
No myofascial pain	33 (11)	160 (53)	193 (64)
Myofascial pain w referral	26 (9)	10 (3)	36 (12)
Myofascial pain wo referral	35 (11)	36 (12)	71 (23)

Table 2. Distribution of self-reported pain sites in the study sample.

	Widespread pain $n = 94$ *n* (%)	No widespread pain ($n = 206$) *n* (%)	Total $n = 300$ *n* (%)
Head/Face	84 (89.4)	78 (37.9)	162 (54)
Neck	85 (90.4)	52 (25.2)	137 (45.7)
Shoulder	70 (74.5)	30 (14.6)	100 (33.3)
Upper arm	72 (76.6)	25 (12.1)	97 (32.3)
Lower arm	44 (46.8)	9 (4.4)	53 (17.7)
Chest	13 (13.8)	2 (1.0)	15 (5.0)
Abdomen	6 (6.4)	5 (2.4)	11 (3.7)
Upper back	62 (66.0)	20 (9.8)	82 (27.3)
Lower back	52 (55.3)	26 (12.6)	78 (26.0)
Hip/ glutaeal area	30 (31.9)	14 (6.8)	44 (14.7)
Upper leg	41 (43.6)	20 (9.8)	61 (20.3)
Lower leg	36 (38.3)	12 (5.8)	48 (16.0)

The population-based weighted prevalence of widespread bodily pain according to the WPI was 15%. The most frequent pain sites outside the orofacial area, were the back, neck, and shoulder/upper arm (Table 2, Figure 2). The overlap between widespread pain and any myofascial orofacial pain diagnosis was 57.3% (Figure 3) and for the subclassifications of myofascial pain with and without referral 72.2% and 49.3%, respectively.

Myofascial pain with and without referral were both associated to widespread pain (OR 4.83 95% CI 2.62–9.05 and 11.62 95% CI 5.18–27.88, respectively; Table 3). Our *post hoc* analysis showed no difference between myofascial pain with or without referral with regards to their association to widespread pain (Table 4).

Discussion

The main finding from this study on myofascial orofacial pain in a community-based sample was the substantial overlap between myofascial pain and widespread pain. Furthermore, the association between myofascial pain and widespread pain is stronger in the presence of pain referral on palpation of the masticatory muscles.

The prevalence of widespread pain in the population varies in the literature, which is likely due to heterogenicity in criteria, methods, samples, and settings. A recent meta-analysis based on 23 papers and 32 prevalence estimates suggested a 10–15% prevalence of widespread pain in the general population [13]. This is in agreement with the population-based weighted prevalence of 15% widespread pain in

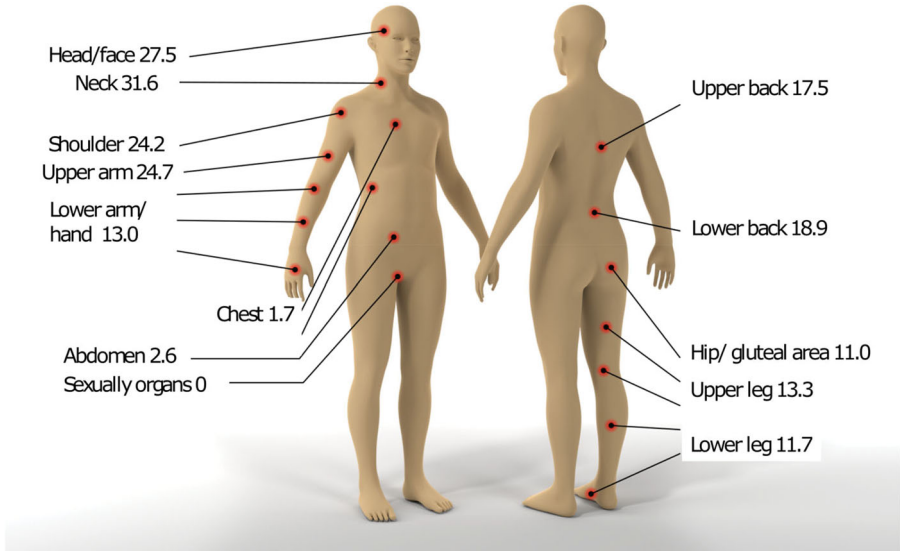

Figure 2. Distribution of self-reported pain sites in the study sample ($n = 300$) together with the population based weighted prevalence (%). Each individual could have reported multiple sites [44].

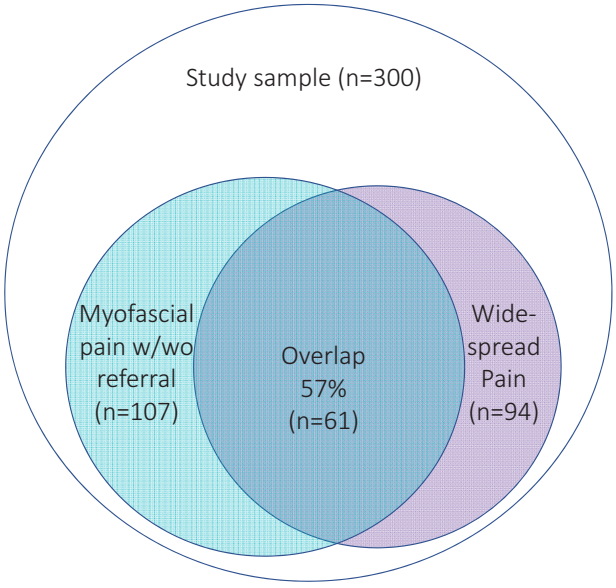

Figure 3. The overlap between myofascial pain (myofascial pain without/with referral) and widespread pain.

Table 3. Associations (Odds Ratio, OR) to widespread pain, when adjusted for the effect of gender and age, respectively.

Variable	OR	95% CI
No myofascial pain (ref)	1	0.02–0.18
Myofascial pain wo referral	4.83	2.62–9.05
Myofascial pain w referral	11.62	5.18–27.88
Female gender	2.21	1.19–4.27
Age	1.02	0.99–1.04

The effect of diagnoses is computed in reference to no myofascial pain.

Table 4. Pairwise comparison based on estimated marginal means of the logistical regression model specified in Table 3.

Variable	OR	95% CI	*p* Value
Myofascial pain w/o referral vs no pain	4.83	2.61–8.96	<0.001
Myofascial pain w referral vs no pain	11.62	5.04–26.77	<0.001
Myofascial pain w/o vs w referral	2.40	0.99–5.80	0.051

Results are averaged over the levels of gender and age.

the community-based sample in the present study. The finding of a higher prevalence of 25% in women is also in accordance with previous findings regarding gender distribution. In contrast, and even though a pronounced overlap between TMD and fibromyalgia in community samples has been reported [16,23], the overlap we found between painful TMD and widespread pain of more than 50% is considerably higher compared to previous reports [22]. This supports the suggestion that TMD is part of a COPC rather than a single symptom [23]. In the present sample, individuals were initially recruited as potential TMD cases based on their answers to three screening questions for TMD [30]. In addition to the presence of pain, the questions also require a frequency of pain, that is, 'once a week or more'. As a consequence, the sample probably included individuals with painful TMD that was more severe when compared to a random general population sample. This in turn might be one of the factors that influenced the substantial overlap with widespread pain. The relationship between pain frequency as one factor in the development and persistence of widespread pain conditions, as indicated by our findings, warrants further studies.

The specific origin of muscle can often be difficult for the patient to pinpoint, especially and in more chronic cases [34]. This clinical feature of muscle pain differs from that of cutaneous pain where is common in a more precise location can often be described. Moreover, any long-lasting pain has been suggested to potentially cause central sensitisation, leading to pain sensations even in areas with no tissue damage. In this respect, referred pain has been suggested to be related to a decrease in pain thresholds to nociceptive input, potentially causing general hyperalgesia and central sensitization [26–28]. However, in an experimental study, Masuda and co-workers have shown that referred pain upon palpation of 1 kg or less was common in healthy individuals [35]. Thus, pain a specific muscle can refer to other muscles and

other tissues such as fascia, and in the orofacial area also to the teeth, even in healthy individuals [26,36]. This contributes to the clinical challenge in precisely determining the origin of the pain. In the DC/TMD, provided that the pain is not better accounted for by another pain diagnosis, the clinician can detect such referred pain from the masticatory muscles according to the criteria for myofascial pain with referral with a sensitivity of 0.86 and specificity of 0.98 [7]. Our results imply that among patients with a myofascial orofacial pain in general practice dentistry, two thirds are likely to also have a widespread pain condition. For individuals who suffer from painful TMD and report concurrent widespread pain, the pain in the orofacial area can probably be interpreted as being related to generalized hyperalgesia and a COPC [23,24,37]. This is also reflected in a previous study from Koutris and co-workers among patients at a specialist clinic, where widespread pain was considered as a confounder for the TMD diagnosis of 'myalgia' [38]. As a consequence of the possible difference in pathophysiology between localised TMD pain and TMD pain as part of a widespread pain condition, standard single treatment with occlusal appliances among individuals with widespread pain may have a poor prognosis, and multimodal and multidisciplinary strategies may instead be required [39]. This is of considerable clinical importance since it was suggested that patients with widespread pain require different treatment strategies when compared to patients with only local pain manifestations [40]. Thus, the therapeutic effect of an occlusal appliance in patients with widespread pain has questionable effect on TMD-pain symptoms and does not outperform instructions given for self-management, such as physical therapy [39,40]. Building on the findings from our study together with previous reports on TMD as a part of a COPC, pain referral in individuals with painful TMD is likely to be associated with a widespread pain condition that warrants further clinical attention. Collectively, increased awareness regarding TMD as a part of an overlapping chronic pain condition can therefore result in improved diagnostics and prognosis along with more effective pain management [41]. The use of pain drawings outside the orofacial area and additional diagnostic methods for widespread pain are of special importance when pain persists or when prior treatments have not been successful [42]. Therefore, in the clinical decision-making for patients with TMD in general dental practice, information on other pain conditions should be included in treatment planning and in the assessment of the expected prognosis.

Previous studies have suggested that pain drawings and self-reported comorbid pains are valid and suitable components of the biopsychosocial profiling when assessing patients with painful TMD [43]. The widespread pain index used in the present study for the definition of widespread pain, with a suggested cut-off at seven or more pain sites, has been shown to be a valid method for the detection of widespread pain [32]. Thus, the validated instruments, methods, and criteria we used, along with a reliable and valid examination procedure, all contribute with self-standing strengths in our study. It is possible that the fact that the examiner was not blinded to the study protocol may have influenced the categorisation of participants with pain and thus have affected the internal validity. Moreover, arthralgia as a potential cause of orofacial pain was not evaluated in this study and should be addressed in future studies on the topic. A potential limitation in the present study is that the study population was recruited primarily for validating the screening questions in relation to the DC/TMD [30]. Since all individuals were invited with focus on TMD and in a random order, without taking the gender of participants into consideration, the study sample had a female dominance. This may, in turn, affect the representativeness of the study population. Nevertheless, the distribution and the weighted prevalence of widespread pain were in line with previous reports, which indicate that the results can be regarded representative for comparable settings.

In conclusion, with a weighted prevalence representative for community samples, our findings reinforce the existing knowledge on the overlap between painful TMD and other chronic pain conditions. Future studies on pain referral as a valid clinical test for central sensitisation are needed.

Disclosure statement

No potential conflict of interest was reported by the authors.

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