# Clinical Findings and Psychosocial Factors in Patients with Atypical Odontalgia: A Case-Control Study

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Dr Thomas List, Orofacial Pain Unit Department of Stomatognathic Physiology, Faculty of Odontology Malmö University, Malmö, Sweden Fax: +46 406658420 E-mail: Thomas.List@od.mah.se Aim: To provide a systematic description of clinical findings and psychosocial factors in patients suffering from atypical odontalgia (AO). Methods: Forty-six consecutive AO patients (7 men and 39 women; mean age, 56 years; range, 31 to 81 years) were compared with 35 control subjects (11 men and 24 women; mean age, 59 years; range, 31 to 79 years). Results: The pain of the AO patients was characterized by persistent, moderate pain intensity (mean,  $5.6 \pm 1.9$ ) with long pain duration (mean,  $7.7 \pm 7.8$  years). Eightythree percent reported that onset of pain occurred in conjunction with dental treatment. No significant difference was found between the groups in number of remaining teeth or number of root fillings. Temporomandibular disorder (TMD) pain (P < .001), tension-type headache (P < .002), and widespread pain (P < .001) were significantly more common among AO patients than controls. Significantly higher scores for somatization (P < .01) and depression (P < .01) and limitations in jaw function (P < .001) were found for the AO group compared with the control group. Significant differences between groups were found in 4 general health domains: role-physical (P < .001), bodily pain (P < .001), vitality (P < .004), and social functioning (P < .001). Conclusion: A majority of the AO patients reported persistent, moderately intense intraoral pain that in most cases had an onset in conjunction with dental treatment. AO patients had more comorbid pain conditions and higher scores for depression and somatization. Significant limitation in jaw function and significantly lower scores on quality of life measures were found for AO patients compared with controls. J OROFAC PAIN 2007;21:89-98

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typical odontalgia (AO) is a chronic pain condition located in the teeth and jaws. Knowledge regarding its etiology, diagnostics, and management continues to be problematic. While other chronic pain conditions in the face, such as temporomandibular disorders (TMD), are abundant and have been well studied epidemiologically, few studies have systematically evaluated AO, and the terminology and specific criteria for classification are still a matter of discussion. AO has been described as a tooth-related pain or pain at a site where a tooth was extracted in absence of clinical and radiographic evidence of tooth pathology or other relevant orofacial hard or soft tissue pathology. 1,2

Some researchers suggest that, in the absence of known etiologic factors, AO is best viewed as an idiopathic pain condition. 1,3-5 Others have pointed out the association between AO and several psychologic factors and therefore emphasized a psychologic origin for the condition.<sup>2,6</sup> However, several reports have proposed that AO is a neuropathic pain condition. Neuropathic pain is a chronic pain condition caused by a lesion or dysfunction of peripheral or central afferent pathways in the nervous system. Accordingly, AO may be viewed as originating from deafferentation of relevant nervous tissue components in the trigeminal system.<sup>2,3,7–9</sup> Nerve injury, which might occur in relation to invasive dental treatment presumed to damage nervous tissue, such as endodontic procedures or tooth extraction, has been reported to be associated with the development of persistent neuropathic orofacial pain. 10 Indeed, animal studies have demonstrated significant changes in secondorder brainstem neurons following deafferentation of the tooth pulp, although in most instances reversal of such post-traumatic neuronal changes has also been reported. 11

Very few studies have been published involving larger samples of AO patients, probably owing to a relatively low prevalence, <sup>12–16</sup> and the lack of available systematic information has yielded, among other things, confusion over how to diagnose and classify AO. The most useful diagnostic methods in patients with orofacial neuropathic pain currently suggested are history taking and somatosensory testing. <sup>17</sup> Somatosensory changes in AO patients, such as hyperesthesia at the pain site, <sup>18,19</sup> allodynia, <sup>5,7,20</sup> exacerbation of pain evoked by temperature, palpation, and percussion have been frequently reported <sup>12,20–22</sup>; however, contradictory findings of no somatosensory abnormalities in atypical facial pain have also been reported. <sup>23</sup>

Neuropathic orofacial pain is reported to be severe and can be accompanied by significant levels of distress.<sup>20</sup> Psychologic factors are important to identify, as they can profoundly influence pain and pain behavior. Pain can disrupt a number of aspects of everyday life, including work, social and recreational activities, and sleep. Because AO is so intimately associated with the teeth and intraoral structures, AO often results in repeated and possibly unnecessary dental measures, such as root canal treatments, apicectomies, and extractions—a vicious circle of treatment visits to numerous health-care professionals.<sup>7</sup> Knowledge of the impact of AO on daily aspects of life compared with healthy controls is lacking.

The present study was part of a larger study<sup>19,24</sup> to address the numerous paradoxes and lack of valid clinical data associated with AO. Its aim was to provide a systematic description of clinical findings and psychosocial factors in patients suffering from AO compared to gender-matched controls. The hypothesis was that psychosocial and behavioral factors and clinical findings in patients with AO do differ significantly from healthy controls and, as an important corollary, that psychosocial and behavioral factors differ significantly more than clinical findings in patients with AO compared with healthy controls.

# **Materials and Methods**

## **Subjects**

Forty-six consecutive AO patients (7 men and 39 women with a mean age of 56 years; range, 31 to 81 years) were compared with 35 control subjects (11 men and 24 women with a mean age of 59 years; range, 31 to 79 years). The AO patients were recruited from orofacial pain clinics in Linköping, Jönköping, Kalmar, and Malmö in Sweden. Age- and gender-matched control patients were recruited from the Public Dental Service clinic in Arlöv, Sweden.

Inclusion Criteria. The AO group had pain located in a region where a tooth had been endodontically or surgically treated, chronic pain of at least 6 months' duration, and pain with no pathological cause detectable in clinical and radiologic examinations. Controls were routine dental patients carefully matched with the AO patients for gender, age, and, as an important feature of the present study, tooth extractions or endodontically treated teeth (trigeminal nerve damage). Furthermore, all controls were free of any acute or persistent orofacial pain complaint.

**Exclusion Criteria**. Patients with a diagnosis of trigeminal neuralgia, herpes zoster, apical periodontitis, maxillary sinusitis, cluster headache, or paroxysmal hemicrania were excluded from both groups. The Regional Ethical Review Board at Linköping University Hospital and the University of Lund approved the study, and all patients signed an informed, written consent. The patients received no monetary compensation.

## **Clinical Measures**

**Pain Location.** Six areas were recorded according to the patients' pain description: Right maxilla, or

1(18) to 5(14); middle maxilla, or 6(13) to 11(23); left maxilla, or 12(24) to 16(28); left mandible, or 17(38) to 21(34); middle mandible, or 22(33) to 27(43); and right mandible, or 28(44) to 32(48).

Clinical Findings. Number of teeth (0 to 32) and number of root fillings (0 to 32) were based upon clinical and radiographic examinations. Mandibular range of motion variables (unassisted opening without pain, maximum unassisted opening) were measured in millimeters. Number of painful sites was based upon the sum of 20 dichotomous (yes/no) muscle palpation pain sites according to the examination that follows the Research Diagnostic Criteria for Temporomandibular Disorders (RDC/TMD) (0 to 20).

## Self-report Measures

Patient Characteristics. Age, gender, and number of therapists visited because of AO pain were documented. The question "Which treatments have been conducted?" (yes/no) was followed by 12 treatment options. The question "What attitude do you have towards pain relieving medication?" was answered on a 0- to 3-point scale; positive, unsure, negative, don't know.

Pain Characteristics. Average pain intensity graded on a 0-to-10 numeric rating scale (NRS), duration of pain (years), and frequency of pain (daily, several times a week, few times a week, 1 to 3 times per month, never) were recorded.

**McGill Pain Questionnaire.** A Swedish version of the short-form McGill Pain Questionnaire (MPQ) was used.<sup>25</sup> It comprised 15 descriptors (11 sensory and 4 affective). Each item was scored on a 0-to-3 point scale (none, mild, moderate, or severe).

Jaw Function Limitation Scale (JFLS). A scale was designed to measure how jaw function is limited during different activities. The scale included 14 items in the domains opening, chewing, communication, and emotions. The patients rated the limitation on a 0-3 point scale. A score of 0 corresponded to no limitation; a score of 3 to extreme limitation.<sup>26</sup>

**Psychologic Status.** Depression and somatization were scored using a shorter version of the Symptom Checklist-90 (SCL-90) according to the RDC/TMD. Twenty of the questions were related to depression and 12 to somatization.<sup>27</sup>

Quality of Life. The generic health-related quality of life measure SF-36, a generic measure of how a person's general health affects their quality of life, was used. The instrument covers 8 domains: physical functioning, role-physical, bodily pain, general

health, vitality, social functioning, role-emotional, and mental health.<sup>28</sup>

Widespread Pain. The patients marked the areas where they have pain on an anatomical drawing. A maximum of 10 areas could be marked: Head, face, mouth (intraoral), throat, neck/shoulder, back, chest, abdomen, upper extremities, and lower extremities.

#### Classification

The RDC/TMD classifies the most common forms of TMD into 3 diagnostic categories and allows multiple diagnoses to be given for a single patient. The RDC/TMD diagnostic categories are myofascial pain; disc displacements; and arthralgia, arthritis, and arthrosis.<sup>27</sup>

Tension-type headache was diagnosed according to the criteria of the International Headache Society (IHS), and patients were assigned 1 of 3 diagnoses: episodic tension-type headache (headache < 15 days/month), chronic tension-type headache (headache > 15 days/month for > 6 months), or no tension-type headache.<sup>29</sup>

#### Design

All patients underwent a dental and radiographic examination and completed the self-report measures. The self-report questionnaire was given to the patients before the examination. Investigators were available to explain the questions if necessary and to check the questionnaires for completeness and legibility. The clinical physical assessment comprised an intraoral evaluation of the teeth and oral mucosa that included inspection, palpation, percussion, electric pulp testing, periodontal probing, and translumination. In addition, radiographic examinations of the jaws and teeth (panorama and periapical radiographs), an examination of the masticatory apparatus according to the RDC/TMD, and a cervical spine examination were made by an experienced orofacial pain specialist (TL) in the AO patients. The controls were examined by a second dentist. Both examiners were calibrated according to the RDC/TMD, and interreliability was checked at 6-month intervals.<sup>30</sup>

#### Statistical Methods

To analyze differences between groups, t tests for independent means were used for continuous variables and the Mann-Whitney test was used for ordinal variables. Chi-square tests were used to assess associations between categorical variables.

Table 1 Pain Characteristics of AO Patients							
	AO group						
Self-report	%	Mean (SD)					
Average pain intensity (NRS)		5.6 (1.9)					
Duration of pain (y)		7.7 (7.1)					
Pain started with dental treatment	83						
No. of pain therapists		4.4 (3.1)					
No. of pain treatments		4.6 (2.8)					
Pain daily or several times a week	87						

NRS = numerical rating scale (0-10).

Fig 1 Distribution of intraoral pain locations. Of the patients, 56% reported pain in the maxilla and 45% reported pain in the mandible. One patient had pain in both the maxilla and the mandible. Some patients had pain in more than 1 area of the jaw.

The Pearson correlation coefficient was used to measure associations between 2 variables on a ratio scale. All inferential statistical tests were 2-tailed and at *P* (5% significance level).

#### Results

## Pain Characteristics

Table 1 summarizes findings for the self-reported pain characteristics measures in the AO group. Fifteen percent of the AO group patients (n = 7)reported their average pain intensity to be 0 to 3 on the NRS; 52% (n = 24) reported a score of 4 to 6, and 33% (n = 15) reported a score of 7 to 10. Fifteen percent reported that the pain began as a toothache. Increased or unchanged pain was reported by 78.3% after rootfilling or extraction of the tooth and by 76.3% after apicectomy. The MPQ pain descriptors used by AO patients, in descending order of frequency, were: tender (78%), throbbing (54%), aching (91%), exhausting (44%), heavy (38%), stabbing (36%), splitting (36%), fearful (33%), burning (31%), sharp (27%), punishing-cruel (24%), gnawing (22%), sickening (18%), cramping (16%), and shooting (13%).

The patients had visited on average 4.4 different therapists for their pain (range 1-20) (Table 1). They received a number of common treatments: occlusal appliances (54%), transcutaneous electrical nerve stimulation (TENS) (43%), occlusal grinding (39%), acupuncture (33%), physical therapy (20%), relaxation (20%), and chiropractic treatment (15%). Overall, 78% of the patients had

used some kind of pharmacologic treatment, and 63% had been treated with physical therapy (including acupuncture and TENS) for their orofacial pain condition. The most commonly used pain-related pharmacologic treatments were analgesics (76%), tranquilizers (31%), sedatives (30%), antidepressants (28%), carbamazepine (18%), and gabapentin (15%). AO patients reported their attitude toward pharmacologic treatment as positive (35%), unsure (33%), negative (15%), and don't know (15%). There was no significant difference in frequency of pain in the left or the right side of the jaw (Fig 1).

A significant correlation was found between depression score and widespread pain (r = 0.44), average pain intensity (r = 39), and worst pain (r = 0.29), whereas no significant correlation was seen between depression score and pain duration (r = -0.15) or number of visited therapists (r = 0.1). A significant correlation was found between somatization score and widespread pain (r = 0.69) and between somatization score and average pain intensity (r = 48), whereas no significant correlation was seen between somatization score and worst pain (r = 0.20), pain duration (r = -0.18), or number of visited therapists (r = 0.18).

#### Comparison of AO and Control Subjects

## Tooth and Masticatory Muscle Clinical Findings.

As Table 2 indicates, no significant difference was found between the 2 groups in number of remaining teeth or number of root fillings revealed by radiography. AO patients had significantly more sites where pain was experienced on palpation of

Table 2 Clinical Findings: Number of Teeth and Root Fillings in AO Patients and Control Subjects

	AO group		Control		
Clinical finding	Mean	SD	Mean	SD	P
No. of teeth	23.5	6.6	23.5	4.1	NS
No. of root fillings	3.4	2.8	2.7	2.1	NS
No. of painful areas upon palpation in the masticatory muscles	5.6	3.9	2.8	3.8	.002

NS = not significant.

Table 3 Comparison of Frequencies of Diagnoses in AO Patients and Control Subjects						
Diagnosis	AO (%)	Control (%)	Р			
RDC/TMD						
Muscle disorder	50.0	2.9	< .001			
Disc displacement	26.1	25.7	NS			
Arthralgia, arthritis, arthrosis	2.2	0	NS			
IHS criteria						
Episodic tension-type headache	45.7	14.7	.002			
Chronic tension-type headache	17.8	0	.002			

Table 4 JFLS and Psychological Status in AO Patients and Control Subjects						
	AC	AO		Control		
Clinical finding	Mean	SD	Mean	SD	P	
JFLS domain						
Chewing	2.4	2.9	0.7	1.9	< .001	
Opening	1.3	2.1	0.3	1.5	< .001	
Communication and emotions	0.8	1.8	0.5	2.5	.046	
Total score Psychological status	4.5	2.3	1.5	5.8	< .001	
Somatization	1.1	0.8	0.6	0.8	.01	
Depression	1.1	8.0	0.78	0.7	.004	

the masticatory muscle (P = .002). Pain drawings revealed significantly more widespread bodily pain (P < .001) in the AO group (mean  $\pm$  SD,  $3.7 \pm 2.7$ ) compared with controls (mean  $\pm$  SD,  $1.3 \pm 2.4$ ). The mean  $\pm$  SD for unassisted opening without pain in the jaw was  $47.5 \pm 6.2$  mm for the AO group and  $49.8 \pm 5.2$  mm for the control group. No significant difference was seen between the groups (P = .08). The mean and SD for maximum unassisted opening of the jaw was  $49.6 \pm 8.3$  mm for the AO group and  $52.4 \pm 5.4$  mm for the control group. No significant difference was seen between the groups (P = .09).

**Pain Diagnoses.** TMD muscle disorder pain diagnoses (RDC/TMD Axis I, Group 1) were significantly more common among AO patients than controls (P < .001), as were the self-reported presence of both episodic and chronic tension-type headaches (P = .002) (Table 3).

**Jaw-Related Disability.** The AO group exhibited significant limitations in jaw function in the domains jaw opening and chewing (P < .001) and communication and emotions (P < .05) compared with the control group, as indicated in Table 4.

**Psychological Variables.** Significantly higher scores on the RDC/TMD Axis II scales for somati-

Table 5 Quality of Life (SF-36) in AO Patients and Control Subjects

	AO		Control		
Domain	Mean	SD	Mean	SD	P
Physical functioning	75.1	24.5	79.8	23.5	.653
Role-physical	44.4	41.6	75.0	41.0	.001
Bodily pain	40.7	20.0	70.6	24.2	< .001
General health	58.8	26.7	70.0	23.3	.059
Vitality	51.2	26.5	67.3	24.2	.004
Social functioning	68.2	28.2	88.2	18.4	.001
Role-emotional	58.0	43.6	75.5	37.9	.068
Mental health	68.1	23.3	77.4	19.8	.072

zation (P = .01) and depression (P = .004) were found in the AO group (Table 4). Distribution of normal, moderate, and severe depression scores were 26%, 26%, and 48% for the AO group and 46%, 37%, and 17% for the controls; distribution values were based upon a population study.<sup>27</sup> Similarly, distribution of normal, moderate, and severe somatization scores were 22%, 28%, and 50% for the AO group versus 51%, 29%, and 20% for the controls. The groups differed significantly in depression (P < .015), and somatization (P < .007).

**Psychosocial Functioning.** Table 5 summarizes SF-36 scores for the AO and control groups. There was a tendency toward a difference between the groups in all the SF-36 domains. However, significant differences between groups were found in 4 SF-36 domains: role-physical (P = .001), bodily pain (P < .001), vitality (P = .004), and social functioning (P = .001).

# Discussion

Overall, the study found that a majority of the AO patients reported persistent, moderately intense intraoral pain that in most cases had an onset in conjunction with dental treatment. TMD pain and tension-type headache were common among AO patients. AO patients, compared to age-matched controls, also revealed significantly more frequent muscle disorder diagnoses and exhibited more widespread bodily pain, significantly higher scores for depression and somatization, and significant limitation of jaw function, and significantly lower quality of life compared with controls.

One major limitation of the present study was the relatively small number of AO cases included, although the sample size was not small compared to most other studies of AO reported. In general, information regarding the prevalence and incidence of AO is limited. For example, even in retrospective studies of endodontic patients, AO has been estimated to be present in approximately 3% to 6% of clinical cases.<sup>31,32</sup>

## **Demographic Issues**

In the majority of published clinical studies investigating AO, the average number of subjects is typically low, which reflects the fact that the condition, although severely painful and disabling, is relatively rare. The present study also encountered this low clinical distribution: To be able to include a sufficient number of participants in the present descriptive study, consecutive patients were pooled from 4 orofacial pain clinics whose demographic composition has been reported as relatively similar. Each clinic is responsible for the population in a large district in Sweden, and together the centers represent a population of approximately 2 million people. The majority of the patients in the present study were women in their late fifties. The gender ratio (more females than males) is similar to the ratios reported for other orofacial pain conditions; however, the age distribution seems to be higher than what is normally seen in TMD studies. 33,34 Since the age and gender distributions in our AO clientele are similar to those reported in other AO studies, the patients studied were considered to be representative of a clinical AO population.<sup>7,16,35</sup>

## Classification Issues

Several classifications and diagnostic criteria have been proposed for AO, but no consensus has yet been reached. 1,2,36,37 Even though the classification systems differ slightly in details, the main case definition characteristics are similar, such as continuous chronic pain in a tooth or at a tooth site and the inability of clinical, radiographic, and laboratory examinations to explain the pathological cause. In the present study, inclusion criteria that fit within these major classification systems were used.5,7,21,38 It has been suggested that several pain conditions may mimic AO and it is important to rule these out before setting a diagnosis of AO.<sup>2</sup> Probably the most difficult diagnoses to rule out are pulpal pain conditions. To exclude these conditions in the present study, a comprehensive anamnesis and intra- and extraoral examinations of the teeth and the masticatory apparatus as well as a radiographic examination were performed.

#### Pain

The most characteristic feature of AO is reported to be intraoral pain that is moderate to severe and persistent.<sup>2,16</sup> In the present study, the vast majority of the patients reported daily or almost daily pain, which is in accordance with other studies. 5,18,20,21,39 Similar findings were reported in 2 studies on neuropathic pain related to facial trauma, maxillofacial surgery, or dental treatment.<sup>22,40</sup> One study found that 81% of the patients reported constant pain, while 19% reported intermittent or only stimulus-evoked pain.<sup>40</sup> In the second study, all the patients exhibited spontaneous continuous pain.<sup>23</sup> Even though the pain is usually continuous, episodes with more acute intense pain have been reported in AO.41 The pain descriptors reported by the AO patients in the present study coincide with reports by others. 7,23,42 However, as Hansson43 has pointed out, commonly used pain descriptors in peripheral or central neuropathic pain have not been identified in the literature, and several descriptors are usually used by a single patient.

AO pain has been reported to be so severe that suicidal attempts have been described. In the present study, the pain intensity ranged from 1 to 10, with an average of 5.6, which reflects a moderate-moderately high pain intensity. The pain intensity in the present study seems be similar to the findings of others. One patient in the present study spoke of suicidal thoughts because of the severity of orofacial pain.

## **Pain Location**

The most common site of AO pain has been found to be the molars and premolars in the maxilla. <sup>5,7,15,21,44</sup> There was also a tendency in the present study for the pain to be more common in the maxilla than the mandible; however, the incisors seemed to be as frequently involved as the premolars and molars. One patient reported pain in both the maxilla and mandible. Vickers and Cousins <sup>36</sup> presented a case where the pain had developed after extraction of the mandibular molars and had spread to the maxilla. Similar changes in regional pain localization to entire parts of the maxilla and mandible have been reported. <sup>5,21,39,45,46</sup>

Approximately two thirds of the patients with presumed neuropathic orofacial pain have been reported to develop TMD.<sup>16</sup> In the present study, the AO patients exhibited significantly more pain in the muscles upon palpation, and 50% were diagnosed as having a TMD muscle disorder.

Headache has been found in several studies to be an accompanying symptom. 47-49 Episodic and chronic tension-type headaches, as well as reported widespread pain, were significantly more common in the AO group. It is interesting to note that pain was more common within the trigeminal and the nontrigeminal innervated areas in the AO group compared with controls. Turp et al<sup>50</sup> found that of 140 consecutive TMD patients, the pain was limited to the face and head in only 29% of the cases. This may indicate that AO is complex and involves both peripheral sensitization and neuronal plasticity of the central and peripheral systems. The findings of frequent pain problems can have implications for the classification of orofacial pain. Indeed, Woda et al<sup>1</sup> suggest that the clinical manifestations of TMD and AO are similar. However, the present study provides additional information that these 2 conditions may also coexist.

#### **Onset of Pain**

A common feature in AO is the report of onset of pain related to dental treatment. In a study of 118 patients who underwent surgical endodontics, Campbell and colleagues<sup>32</sup> found that 6 (5%) had persistent pain following surgery, 3 had had pain before surgery and experienced no postoperative pain reduction, and 3 developed pain following surgery. In the present study, a majority reported onset of pain following endodontic measures, apicectomies, or extractions of teeth, which is in line with the findings in others. 10,16 Most of the patients in the present study who did not report dental treatment as the main cause for the onset of pain instead reported initial toothache for which they were seeking treatment. A majority of the patients reported increased or unchanged pain intensity after dental and surgical treatment. However, as pointed out by Vickers and Cousins,<sup>36</sup> it is impossible at this stage to state categorically that dental/endodontic treatment is a prime causal factor in the development of neuropathic pain. While various endodontic procedures inflict mechanical and chemical trauma on the pulpal and periodontal nociceptors, pre-existing pulpitis may well be a trigger for neuropathic pain. Mogil and Grisel<sup>51</sup> reported that postoperative neuropathic pain is more prevalent following amputation when there is concurrent pain and when there has been severe postoperative pain. A similar finding was seen in a 1-year follow-up study of 175 patients undergoing endodontic treatment where chronic pain, pain intensity, and previous episodes of painful treatments were associated with the development of chronic pain.<sup>52</sup>

There have been case reports of patients where all teeth had undergone root canal treatment and apicectomies.8,20,31,38 Vickers and Cousins36 reported that 1 patient who had 5 teeth extracted required more tooth extractions because the pain remained unchanged. In the present study sample, however, the average number of root canal fillings and number of remaining teeth did not differ from the controls. Possible explanations for this could be a good dissemination of knowledge within the dental community combined with a high level of education in addition to high-quality preventive dentistry. This finding at least indicates that cases where excessive dental treatment has been conducted are rare, and it may be that in these individuals the pain behavior differs and is related more to personality traits than to pain.

## **Treatment Seeking**

In general, neuropathic pain is considered to have a lower rate of treatment success than nociceptive pain. This would invite patients to increase treatment seeking and "doctor shopping." Pfaffenrath et al<sup>42</sup> reported that patients with atypical facial pain had consulted an average of 7.5 professionals, including dentists; general medical practitioners; neurologists; ear, nose, and throat specialists; maxillofacial surgeons; psychiatrists; ophthalmologists; and dermatologists. In the present study, approximately 4 different therapists had been consulted on average. This seems low in light of the persistence of their pain; average duration of pain was approximately 8 years. A low frequency in the use of antidepressants and antiepileptic drugs compared with a variety of other treatment modes was found in the present study. Since these drugs have been found to reduce pain in neuropathic and chronic pain conditions, an increase in the use of these drugs might be an option for AO patients to reduce their suffering.<sup>9,53</sup> However, few randomized clinical trials have been conducted to evaluate the pain-relieving effect of pharmacologic interventions in chronic orofacial pain, and the scientific evidence for their efficacy is weak.53

## **Psychologic Status**

Several studies have supported an association between AO and different psychological conditions such as depression, 8,12,14,41,54 somatoform pain disorder, 38,41,55 and anxiety. 44 In the present study, a significant difference was seen between AO patients and controls in depression and somatization scores, with 48% of the AO patients

exhibiting severe depression scores and 50% exhibiting severe somatization scores. The elevated scores for depression in the present study are approximately in line with the findings of other studies. 12,14,56 Graff-Radford and Solberg 56 used the Minnesota Multiphasic Personality Inventory (MMPI) to evaluate patients with AO and compared the results with those of headache patients. They found essentially no difference in the profiles of the conditions; however, this could be related to psychometric properties of the MMPI. Overall, this may suggest that psychologic factors are important but that they do not differ dramatically compared to other pain conditions. Recent literature has emphasized a multidimensional concept of pain in which several axes are used.<sup>27</sup> In the RDC/TMD, Axis I corresponds to physical measures and Axis II to psychosocial status and distress. This 2-axis approach has been found to be useful in the diagnosis and management of TMD and has been recommended as a model for other pain conditions.<sup>57</sup> It may also be adaptable to be similarly useful for characterizing AO patients.

## Quality of Life

Generic instruments such as the SF-36 cover a broad spectrum of domains to encompass different aspects of quality of life. In almost all domains, AO patients exhibited a significant reduction or a tendency toward a reduction in quality of life compared to the controls. The greatest impact was on the pain-related domains and social functioning. Studies focusing on pain behavior have shown that pain has an impact on several aspects of social functioning and daily activities. In the present study every domain measured by the SF-36 showed a greater impact on the AO group compared to the control group; those domains where there were no differences in SF-36 between AO and control groups were probably a function of the relatively small sample size, which may not have allowed sufficient statistical power to reveal statistical differences in those domains. However, it is unlikely that there would be a difference between the groups in the physical functioning domain, since this domain reflects physical activities such as climbing stairs and carrying a bag of groceries, which are not influenced by intraoral pain such as AO.

To supplement the generic instrument, the JFLS was included to evaluate the limitation in masticatory function. In a previous study, the scale found a significant limitation in TMD patients compared with controls.<sup>58</sup> In the present study, the AO

patients exhibited a jaw limitation compared with the controls. The subjective limitation in jaw opening could, however, not be confirmed with the clinical measurement of the jaw-opening capacity. A possible explanation may be methodological differences in that the JFLS reflects jaw function related to daily tasks such as opening wide enough to bite from a whole apple, whereas the clinical measurement of the jaw opening is a 1-time registration following an instruction. It has been reported that some AO patients have had difficulty chewing with full or partial dentures owing to increased sensitivity in the mucosa. 40 It should, however, be emphasized that in the present study the orofacial limitation was limited not only to chewing but also to other aspects such as opening the mouth wide, communicating, and expressing facial emotions.

# **Conclusions**

AO was more frequent among women than men. A majority of the AO patients reported persistent, moderately intense intraoral pain that in most cases had an onset in conjunction with dental treatment. TMD pain and tension-type headache were common among the AO patients. The AO patients had more widespread bodily pain, higher scores for depression and somatization, and exhibited a significant limitation in jaw function and quality of life compared with controls. It is abundantly clear that the major lack of systematic scientific data with regard to this important chronic orofacial pain condition necessitates better and more inclusive studies. Future multicenter as well as multidisciplinary research will be needed in order to develop sufficient clinical sample sizes for more systematic and comprehensive research as well as comparisons with other orofacial pain conditions.

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## References

- 1. Woda A, Tubert-Jeannin S, Bouhassira D, et al. Toward a new taxonomy of idiopathic orofacial pain. Pain 2005:116:396–406.
- 2. Melis M, Lobo SL, Ceneviz C, et al. Atypical odontalgia: A review of the literature. Headache 2003;43:1060–1074.
- Woda A, Pionchon P. Orofacial idiopathic pain: Clinical signs, causes and mechanisms [in French]. Rev Neurol (Paris) 2001;157:265–283.
- Woda A, Pionchon P. A unified concept of idiopathic orofacial pain: Pathophysiologic features. J Orofac Pain 2000;14:196–212.
- Woda A, Pionchon P. A unified concept of idiopathic orofacial pain: Clinical features. J Orofac Pain 1999;13:172–184.
- Feinmann C, Newton-John T. Psychiatric and psychological management considerations associated with nerve damage and neuropathic trigeminal pain. J Orofac Pain 2004;18:360–365.
- Graff-Radford SB, Solberg WK. Atypical odontalgia. J Craniomandib Disord 1992;6:260–265.
- Marbach JJ. Is phantom tooth pain a deafferentation (neuropathic) syndrome? Part I: Evidence derived from pathophysiology and treatment. Oral Surg Oral Med Oral Pathol 1993;75:95–105.
- 9. Jensen TS. An improved understanding of neuropathic pain. Eur J Pain 2002;6:3–11.
- Lynch ME, Elgeneidy AK. The role of sympathetic activity in neuropathic orofacial pain. J Orofac Pain 1996;10:297–305.
- 11. Sessle BJ. Acute and chronic craniofacial pain: Brainstem mechanisms of nociceptive transmission and neuroplasticity, and their clinical correlates. Crit Rev Oral Biol Med 2000;11:57–91.
- 12. Rees RT, Harris M. Atypical odontalgia. Br J Oral Surg 1979;16:212–218.
- 13. Marbach JJ. Phantom tooth pain. J Endod 1978;4:362-372.
- Brooke RI. Atypical odontalgia. A report of twenty-two cases. Oral Surg Oral Med Oral Pathol 1980;49:196–199.
- Pollmann L. Determining factors of the phantom tooth. N Y State Dent J 1993;59:42–45.
- Vickers ER, Cousins MJ, Walker S, Chisholm K. Analysis of 50 patients with atypical odontalgia. A preliminary report on pharmacological procedures for diagnosis and treatment. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1998;85:24–32.
- 17. Svensson P, Baad-Hansen L, Thygesen T, Juhl GI, Jensen TS. Overview on tools and methods to assess neuropathic trigeminal pain. J Orofac Pain 2004;18:332–338.
- 18. Gross SG. Atypical odontalgia: A cause for dental failure. J Conn State Dent Assoc 1991;67:36, 37.
- 19. Baad-Hansen L, List T, Jensen TS, Svensson P. Increased pain sensitivity to intraoral capsaicin in patients with atypical odontalgia. J Orofac Pain 2006;20:107–114.
- Marbach JJ. Orofacial phantom pain: Theory and phenomenology. J Am Dent Assoc 1996;127:221–229.
- Pertes RA, Bailey DR, Milone AS. Atypical odontalgia—A nondental toothache. J N J Dent Assoc 1995;66:29–31, 33.
- 22. Eide PK, Rabben T. Trigeminal neuropathic pain: Pathophysiological mechanisms examined by quantitative assessment of abnormal pain and sensory perception. Neurosurgery 1998;43:1103–1110.

- 23. Lang E, Kaltenhauser M, Seidler S, Mattenklodt P, Neundorfer B. Persistent idiopathic facial pain exists independent of somatosensory input from the painful region: Findings from quantitative sensory functions and somatotopy of the primary somatosensory cortex. Pain 2005;118:80-91.
- 24. Baad-Hansen L, List T, Jensen TS, Leijon G, Svensson P. Blink reflexes in patients with atypical odontalgia. J Orofac Pain 2005;19:239-247.
- 25. Burckhardt CS, Bjelle A. A Swedish version of the shortform McGill Pain Questionnaire. Scand J Rheumatol 1994;23:77-81.
- 26. Ohrbach R, List T. Psychometric properties of the jaw functional limitation scale [abstract 1023]. J Dent Res 2002:147.
- 27. Dworkin SF, LeResche L. Research Diagnostic Criteria for Temporomandibular Disorders: Review, criteria, examinations and specifications, critique. J Craniomandib Disord 1992;6:301-355.
- Ware JE Jr, Sherbourne CD. The MOS 36-item short-form health survey (SF-36). I. Conceptual framework and item selection. Med Care 1992;30:473-483.
- 29. Headache Classification Subcommittee of the International Headache Society. The International Classification of Headache Disorders: 2nd edition. Cephalalgia. 2004;24(suppl 1):9-160.
- 30. John MT, Dworkin SF, Mancl LA. Reliability of clinical temporomandibular disorder diagnoses. Pain 2005;118:61-69.
- 31. Marbach JJ, Hulbrock J, Hohn C, Segal AG. Incidence of phantom tooth pain: An atypical facial neuralgia. Oral Surg Oral Med Oral Pathol 1982;53:190-193.
- 32. Campbell RL, Parks KW, Dodds RN. Chronic facial pain associated with endodontic therapy. Oral Surg Oral Med Oral Pathol 1990;69:287-290.
- 33. Dworkin SF, Huggins KH, LeResche L, et al. Epidemiology of signs and symptoms in temporomandibular disorders: Clinical signs in cases and controls. J Am Dent Assoc 1990;120:273-281.
- 34. List T, Dworkin SF. Comparing TMD diagnoses and clinical findings at Swedish and US TMD centers using research diagnostic criteria for temporomandibular disorders. J Orofac Pain 1996;10:240-253.
- 35. Schnurr RF, Brooke RI. Atypical odontalgia. Update and comment on long-term follow-up. Oral Surg Oral Med Oral Pathol 1992;73:445-448.
- 36. Vickers ER, Cousins MJ. Neuropathic orofacial pain part 1—Prevalence and pathophysiology. Aust Endod J 2000;26:19-26.
- 37. Zakrzewska JM. Classification issues related to neuropathic trigeminal pain. J Orofac Pain 2004;18:325-331.
- Marbach JJ, Raphael KG. Phantom tooth pain: A new look at an old dilemma. Pain Med 2000;1:68-77.
- 39. Biron CR. Atypical odontalgia is often dismissed as "vivid imagination" during diagnosis. RDH 1996;16(9):40-42,

- 40. Gregg JM. Studies of traumatic neuralgia in the maxillofacial region: Symptom complexes and response to microsurgery. J Oral Maxillofac Surg 1990;48:135-140.
- 41. Marbach JJ. Phantom tooth pain: Differential diagnosis and treatment. N Y State Dent J 1993;59:28-33.
- 42. 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.
- 43. Hansson P. Neuropathic pain: Clinical characteristics and diagnostic workup. Eur J Pain 2002;6(suppl A):47-50.
- 44. Klausner JJ. Epidemiology of chronic facial pain: Diagnostic usefulness in patient care. J Am Dent Assoc 1994;125:1604-1611.
- 45. Bates RE Jr, Stewart CM. Atypical odontalgia: Phantom tooth pain. Oral Surg Oral Med Oral Pathol 1991;72:479-483.
- 46. Goss AN, McNamara J, Rounsefell B. Dental patients in a general pain clinic. Oral Surg Oral Med Oral Pathol 1988;65:663-667.
- 47. Nicolodi M, Sicuteri F. Phantom tooth diagnosis and an anamnestic focus on headache. N Y State Dent J 1993;59:35-37.
- 48. Sicuteri F, Nicolodi M, Fusco BM, Orlando S. Idiopathic headache as a possible risk factor for phantom tooth pain. Headache 1991;31:577-581.
- 49. Reik L Jr. Atypical odontalgia: A localized form of atypical facial pain. Headache 1984;24:222-224.
- 50. Turp JC, Kowalski CJ, Stohler CS. Temporomandibular disorders-Pain outside the head and face is rarely acknowledged in the chief complaint. J Prosthet Dent 1997;78:592-595.
- 51. Mogil JS, Grisel JE. Transgenic studies of pain. Pain 1998;77:107-128.
- 52. Polycarpou N, Ng YL, Canavan D, Moles DR, Gulabivala K. Prevalence of persistent pain after endodontic treatment and factors affecting its occurrence in cases with complete radiographic healing. Int Endod J 2005;38:169-178.
- 53. List T, Axelsson S, Leijon G. Pharmacologic interventions in the treatment of temporomandibular disorders, atypical facial pain, and burning mouth syndrome. A qualitative systematic review. J Orofac Pain 2003;17:301-310.
- 54. Kreisberg MK. Atypical odontalgia: Differential diagnosis and treatment. J Am Dent Assoc 1982;104:852-854.
- 55. Remick RA, Blasberg B, Barton JS, Campos PE, Miles JE. Ineffective dental and surgical treatment associated with atypical facial pain. Oral Surg Oral Med Oral Pathol 1983;55:355-358.
- 56. Graff-Radford SB, Solberg WK. Is atypical odontalgia a psychological problem? Oral Surg Oral Med Oral Pathol 1993;75:579-582.
- 57. Garofalo J, Wesley A. Research Diagnostic Criteria for Temporomandibular Disorders: Reflection of the physicalpsychological interface. APS Bulletin 1997;7(3):4-16.
- List T, Paulin G, Lundström I, Ohrbach R. Orofacial disorder diagnosis: Relationship to jaw limitation scale [abstract 1024]. J Dent Res 2002:148.