CLINICAL STUDY OF PATIENTS WITH PERSISTENT OROFACIAL PAIN

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ABSTRACT - Objective: To evaluate a sample of patients with persistent facial pain unresponsive to prior treatments.

Methods: Hospital records of 26 patients with persistent facial pain were reviewed (20 female and 6 male).

Results: Patients were classified into three groups according to their presenting symptoms: a) Group I, eight patients (30.7%) with severe, diffuse pain at the face, teeth or head; b) Group II, eight patients (30.7%) with chronic non-myofascial pain and; c) Group III, ten patients with chronic myofascial pain (38.4%). We find 11 different diagnoses among the 26 patients: pulpitis(7), leukemia(1), oropharyngeal tumor(1), atypical odontalgia(1), Eagle's syndrome(1), trigeminal neuralgia(4), continuous neuralgia(1), temporomandibular disorders(9), fibromyalgia(2), tension-type headache(1), conversion hysteria(2). After the treatment program all patients had a six-month follow-up period with pain relief, except the patient with tumor.

Conclusion: The wide variability of orofacial pain diagnosis (benign to life-threatening diseases) indicates the necessity to reevaluate patients presenting recurrent pain that is refractory to the usual treatments.

KEY WORDS: orofacial pain, trigeminal neuralgia, tumor, temporomandibular disorders, atypical facial pain

Estudo clínico de pacientes com dor orofacial persistente

RESUMO - Objetivo: Avaliar uma amostra de doentes com dor facial persistente.

Método: Foram revisados 26 prontuários de doentes com dor facial persistente (20 mulheres e 6 homens).

Resultados: Classificação dos doentes, após o diagnóstico: a) Grupo I, oito pacientes (30,7%) com dor facial com forte intensidade; b) Grupo II, oito pacientes (30,7%) com dor crônica de natureza não-miofascial e; c) Grupo III, dez pacientes com dor crônica miofascial (38,4%). Foram encontrados 11 diagnósticos diferentes entre os 26 pacientes: pulpites(7), leucemia(1), tumor de orofaringe(1), odontalgia atípica(1), síndrome de Eagle(1), neuralgia idiopática do trigêmeo(4), neuralgia atípica(1), disordens temporomandibular(9), fibromyalgia(2) cefaléia tipo-tensão(1), histeria de conversão(2). O acompanhamento dos doentes, após receberem a respectiva terapia, foi de seis meses, com alívio da dor, exceto para o doente com tumor de orofaringe.

Conclusão: A variabilidade das fontes da dor facial inclui doenças benignas e doenças graves, sendo indispensável a reavaliação de doentes que não respondem aos tratamentos convencionais para a dor.

PALAVRAS-CHAVE: dor orofacial, neuralgia trigeminal, tumor, articulação temporomandibular, dor facial atípica.

The complex innervation and function of facial structures makes the diagnosis of facial pain and its treatment very difficult and frustrating1,2. Patients with chronic facial pain, even after receiving multiple treatments, should be carefully reassessed and clinically re-examined. Myofacial pain syndromes, temporomandibular disorders (TMD), neuralgias, ENT diseases, dental pain, tumors, neurovascular pain or psychiatric diseases frequently present with overlapping signs and symptoms3,4. Referred, severe, acute pain frequently makes the diagnosis difficult.

It is known that dental pain can radiate to the face and simulate other sources of pain due to sensitization of neurons in the central nervous system5,6. This can cause other phenomena including...
secondary adjacent muscle hyperactivity. Patients can present with muscular hyperactivity resulting from persistent acute or chronic pain, and elicit secondary muscle pain. Therefore, the elimination of the primary source of pain is essential but, in chronic pain, is not always enough for control of pain. An incorrect and ineffective treatment may perpetuate or generate chronic pain. The understanding of secondary pain mechanisms of craniofacial pain is necessary for the formulation of an accurate diagnosis.

The goal of this study is to demonstrate the variability of possible diagnoses in a sample of patients with persistent facial pain and how the orofacial pain specialty team was able to assist in their diagnosis and management.

**METHOD**

This is a retrospective study of a sample of patients with persistent facial pain unresponsive to previous treatments who were referred to an orofacial pain team of a large teaching hospital. The study was approved by the Ethics Commission of the hospital.

The records of 26 consecutive patients, 20 female and 6 male, were analyzed. Patients were selected according to the description of their chronic pain symptoms and classified in accordance with the criteria of the International Association for the Study of Pain and references of the American Academy of Orofacial Pain. This sample represents 4% of all new patients with orofacial pain complaints admitted to the Orofacial Pain Clinic during the period of this study (August 1992 - December 1996). In order to ensure consistency in interview methods, the diagnoses were confirmed by clinical examination performed by members of a trained and calibrated interdisciplinary pain team.

The standardized diagnostic protocol was applied to all patients equally. It consists of a standardized interview and systematic evaluation of cervical, cranial, facial, oral and dental structures. The general characteristics for differential diagnosis of the diseases are presented in Table 1. Diagnoses were:

- Pulpitis – History of daily, throbbing, diffuse pain, trigger by hot or cold and waking the patient during the night.

**Table 1. Differential diagnosis in pain frequently observed in the orofacial region.**

<table>
<thead>
<tr>
<th>Disease</th>
<th>Idiopathic trigeminal neuralgia</th>
<th>Pulpitis (referred dental pain)</th>
<th>Orofacial neoplasia</th>
<th>Temporomandibular disorders</th>
<th>Fibromyalgia</th>
<th>Eagle's syndrome</th>
</tr>
</thead>
<tbody>
<tr>
<td>Pain</td>
<td>Electric shock-like</td>
<td>Trobbing</td>
<td>Variable (atypical)</td>
<td>Dull, stabbing</td>
<td>Dull</td>
<td>Dull</td>
</tr>
<tr>
<td>Duration</td>
<td>Seconds</td>
<td>Minutes to hours</td>
<td>Variable</td>
<td>Minute to hours</td>
<td>Constant</td>
<td>Short-duration</td>
</tr>
<tr>
<td>Intensity</td>
<td>Severe</td>
<td>Sight to severe</td>
<td>Severe</td>
<td>Moderate</td>
<td>Moderate</td>
<td>Middle</td>
</tr>
<tr>
<td>Localization</td>
<td>Good</td>
<td>Diffuse</td>
<td>Diffuse</td>
<td>Good, Diffuse</td>
<td>Diffuse</td>
<td>Diffuse</td>
</tr>
<tr>
<td>Characteristics</td>
<td>Trigger zone, diurnal</td>
<td>Diurnal or nocturnal dental sensitivity, look for dental problem</td>
<td>Referred pain frequently neurological signs, WBC abnormalities</td>
<td>TMJ or musle, pain to movement, limited open mouth</td>
<td>Generalized body pain, spontaneous</td>
<td>Usually pain is in the throat / mouth floor</td>
</tr>
<tr>
<td>Local treatment</td>
<td>No</td>
<td>Dental treatments, local anesthesia blocks the pain</td>
<td>Surgical</td>
<td>Physical therapy, splints, anesthetic injection,</td>
<td>Physical therapy</td>
<td>Corticoid injection, surgery</td>
</tr>
<tr>
<td>General treatment</td>
<td>Anti-convulsivants</td>
<td>NSAIDs, analgesics</td>
<td>Chemotherapy, radiotherapy HSCT</td>
<td>NSAIDs</td>
<td>TAD, myorelaxants</td>
<td>NSAIDs</td>
</tr>
<tr>
<td>Trigger</td>
<td>Non-noxious stimulus</td>
<td>Mechanical, foods, cold, heat, suit</td>
<td>Jaw movement</td>
<td>Palpation, jaw function</td>
<td>Palpation, function</td>
<td>Swallowing</td>
</tr>
</tbody>
</table>

TMJ, temporomandibular joint; NSAIDs, nonsteroidal antiinflammatory drugs; TAD, tricyclic antidepressants; WBC, white blood count; HSCT, hematopoietic stem cells transplantation.
Table 2. General characteristics of the sample (n = 26).

<table>
<thead>
<tr>
<th>P</th>
<th>A</th>
<th>G</th>
<th>N</th>
<th>Pain side</th>
<th>Pain intensity</th>
<th>Pain duration (months)</th>
<th>Descriptive terms (before the final diagnosis)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>13</td>
<td>M</td>
<td>4</td>
<td>L</td>
<td>M/S</td>
<td>3</td>
<td>Dental and TMJ constant pain, generalized dental mobility, paresthesia of the left lower lip.</td>
</tr>
<tr>
<td>2</td>
<td>35</td>
<td>F</td>
<td>3</td>
<td>R</td>
<td>S</td>
<td>30</td>
<td>Jaw and facial throbbing pain; diurnal/nocturnal; dental generalized sensitivity to percussion.</td>
</tr>
<tr>
<td>3</td>
<td>41</td>
<td>F</td>
<td>4</td>
<td>R</td>
<td>S</td>
<td>10</td>
<td>Dental and facial pulsating pain; headache; diurnal/nocturnal; dental sensitivity to percussion of #17; cold water provoked the severe attack.</td>
</tr>
<tr>
<td>4</td>
<td>39</td>
<td>F</td>
<td>5</td>
<td>L</td>
<td>S</td>
<td>60</td>
<td>Dental and facial constant pain; diurnal/nocturnal; NSAIDs, TAD, splint, antibiotics, root therapy (teeth: 33,34,35), tooth extraction (#35) without relief of the pain; she had been a 3 years history of TMD that was controlled totally; tenderness of the masticatory muscles.</td>
</tr>
<tr>
<td>5</td>
<td>60</td>
<td>M</td>
<td>2</td>
<td>R</td>
<td>S</td>
<td>10</td>
<td>Headache and dental constant pain; diurnal/nocturnal; pain beginning during flying; presence of subgingival decay at distal face of tooth #13. The pain was referred to superior teeth and temporal region. An attack was caused by air flow of the dental equipment into the dental cavity (#13).</td>
</tr>
<tr>
<td>6</td>
<td>50</td>
<td>F</td>
<td>2</td>
<td>L</td>
<td>S</td>
<td>20</td>
<td>Facial and dental pulsating pain; paresthesia of the left lower lips; history of the jaw fracture 1 year before this complaint.</td>
</tr>
<tr>
<td>7</td>
<td>40</td>
<td>F</td>
<td>3</td>
<td>R</td>
<td>S</td>
<td>30</td>
<td>Facial jabbing pain; diurnal/nocturnal; subgingival decay at the tooth #18.</td>
</tr>
<tr>
<td>8</td>
<td>31</td>
<td>M</td>
<td>5</td>
<td>R</td>
<td>S</td>
<td>7</td>
<td>Cervical and facial jabbing pain; diurnal/nocturnal; NSAIDs, TAD, benzodiazepines, codeine and physical cervical therapy without relief of the pain; he had been one day hospitalization due the intensity of the pain.</td>
</tr>
<tr>
<td>9</td>
<td>22</td>
<td>M</td>
<td>15</td>
<td>L</td>
<td>S</td>
<td>12</td>
<td>Left TMJ pulsating pain, diurnal/nocturnal, complete dentition, dislocation of the left TMJ disc, painful jaw movement.</td>
</tr>
<tr>
<td>10</td>
<td>18</td>
<td>F</td>
<td>3</td>
<td>R</td>
<td>M</td>
<td>24</td>
<td>Constant dental and facial pain, tooth extraction of #18 without relief of the pain, history of chronic throat pain.</td>
</tr>
<tr>
<td>11</td>
<td>55</td>
<td>F</td>
<td>3</td>
<td>L</td>
<td>M</td>
<td>24</td>
<td>Jaw episodic pain, cancerophobia</td>
</tr>
<tr>
<td>12</td>
<td>35</td>
<td>F</td>
<td>7</td>
<td>B</td>
<td>M</td>
<td>72</td>
<td>TMJ constant pain; had been carried out 4 TMJ surgery, multiple tooth extraction, complete denture, physical, and pharmacological treatments without any relief of the pain. Epilepsy.</td>
</tr>
<tr>
<td>13</td>
<td>38</td>
<td>F</td>
<td>3</td>
<td>L</td>
<td>S</td>
<td>6</td>
<td>Facial electric shock-like episodic pain and dull pain; during the first appointment she had 15 attacks lasting from 15 to 40 seconds each one. Masseter muscle tenderness.</td>
</tr>
<tr>
<td>14</td>
<td>40</td>
<td>F</td>
<td>1</td>
<td>B</td>
<td>M</td>
<td>60</td>
<td>TMJ constant pain; oral rehabilitation procedures, dental splints and physical therapy with improvement of jaw movement, without relief of the pain. Pain started after a spinal anesthesia.</td>
</tr>
</tbody>
</table>

Continues
night. The dental source of pain was identified by a clinical evaluation and diagnostic anesthetic block12.

Acute leukemia – Diagnosis is made with laboratory examination of the peripheral blood (WBC) and bone marrow. WBC is usually elevated, but some case present with normal or decreased counts. Anemia (pallor, shortness of breath and fatigue) and thrombocytopenia are other frequent clinical findings13. Oral signs and symptoms frequently lead to a diagnosis of the leukemia; i.e., looseness and mobility of the teeth and paresthesia of the lips is reported during the leukemic cellular infiltration in the periodontal membrane and peripheral trigeminal nerve, respectively13.

Trigeminal neuralgia – History and clinical evaluation
and the presence of paroxysmal and electric shock-like pain with a trigger zone.

Tumor (Oropharyngeal) – Based on computed tomography (CT) and magnetic resonance image (MRI) of the cranial and facial region. The clinical characteristics of the pain are variable and atypical.

Temporomandibular disorders – The diagnosis is by history and clinical exam. Inclusionary criteria include the presence of limited opening, tenderness of the masticatory muscles and joint sounds during mandibular function. CT is used in degenerative process of the temporomandibular joint (TMJ).

Eagle syndrome – Characterized by pain in the oropharyngeal region during mandibular activities, mainly swallowing. Image exams usually show a styloid process elongated and the inflammation is the cause of the pain. Palpation of the posterior and medial region of the mandible angle is painful. The precise diagnosis is made with clinical examination.

Fibromyalgia – Characterized by widespread pain, decreased pain threshold, sleep disturbance, fatigue, psychological distress and chronic headache. Patients thought to have fibromyalgia were diagnosed based on the demonstration of multiple tender points. These tender points were bilaterally, symmetrical but did not refer pain when provoked. Diagnosis in this condition is clinical.

Diagnosis of mental health disorders was made by a psychiatric examination according to the diagnostic criteria for hysterical conversion, or pain associated with depression.

Radiographic and laboratory evaluation - Panoramic radiography of the jaw was performed for all patients. CT scan of the craniofacial region with contrast, MRI and hematological examination (complete blood count) was performed in those cases with recurrent pain without clinical evidences of benign pain if a structural lesion was suspected. CT, MRI and hematological tests were made for patients with clinical diagnostic of trigeminal neuralgia for differential diagnostic between idiopathic and symptomatic trigeminal neuralgia.

Treatment – Patients received appropriate treatment after achieving an accurate diagnosis. Pulpitis was treated with conventional dental management. Trigeminal neuralgia was treated with carbamazepine. Patients with systemic disorders, oropharyngeal tumor and leukemia, were referred for specific treatment, according to the diagnosis. No further commentary regarding treatment of systemic diseases is included in this study as these patients were referred out of the study. A pain assessment was performed immediately after the treatment program and after a six-month follow-up period. A subjective scale was used for this evaluation included five items: SD (Pain free), O (Optimum), S (Satisfactory), PM (Poor improvement) and SM (Without improvement).

Statistical analysis – Patients were separated into groups and comparisons were conducted for general characteristics of the sample. The data was analyzed in the SPSS 10 for Windows program.

RESULTS

In 80.7% of the patients the previous diagnosis was incorrect and the average number of health professionals consulted was 4.7.

Patients were classified into three groups according to their final diagnoses: Group I - Acute pain: Eight patients (30.7%) with severe, diffuse pain referred to the ipsilateral face, teeth or temporal region;

Group II - Chronic non-myofascial pain: Eight patients (30.7%) with chronic non-myofascial pain and;

Group III - Chronic myofascial pain: Ten patients (30.7%) with chronic myofascial pain (38.4%) associated with other painful comorbidities such as fibromyalgia, trigeminal neuralgia and continuous neuralgia.

Acute and chronic pain conditions were classified according to the IASP classification of chronic pain. Throbbing pain during the day or the night was more common in dental lesions; paroxysmal pain was the most common expression of trigeminal neuralgias; and constant, pressure and burning pain were the most common complaints in chronic musculoskeletal pain.

Group I - Acute pain – Eight patients (5 women and 3 men) in otherwise good oral health, aging 13 to 60 years old (median 39.50±13.74) described diffuse and severe pain during the day and at night with pain referring to the facial or cranial region. On average, pain was of one-month duration. Prior diagnoses were trigeminal neuralgia, TMD, myofascial pain, and atypical facial pain. A re-evaluation led to the correct diagnosis of leukemia in one patient and pulpitis in seven others. Pulpitis arose in the following teeth: an inferior canine (#43), two inferior pre-molars (#34, #35), two superior molars (#17, #18) and two inferior molars (#46, #47). Two patients reported referred pain to the adjacent teeth and to ipsilateral face; one patient had referred pain to all ipsilateral teeth, face and temporal area; one patient had referred pain of the opposing teeth, ipsilateral face and temporal area; and three patients had referred pain to the ipsilateral face. In six patients, the teeth were painful to vertical and/or horizontal percussion and in three, airflow stimulation of the teeth trigged the pain. Pulpitis was treated with conventional dental therapy. The patient with leukemia was
referred to a hematologist and received chemotherapy and a bone marrow transplant.

In summary, in the Group I, seven patients presented with diffuse unilateral craniofacial pain from dental causes (pulpitis), and one patient had dental pain and general dental mobility because of a systemic disorder. Two patients in this group received either dental extractions or oral surgery for the treatment without any improvement prior to receiving a final diagnosis.

All the patients of this group were pain free after an accurate diagnosis and correct treatment and remained so at six months of follow-up evaluation.

Group II - Chronic non-myofascial pain - Eight patients (3 men and 5 women), ranging in age from 18 to 57 years old (median 36.50±13.59) reported unilateral (6) and bilateral (2) localized pain which was diurnal and moderate to severe in the facial or cranial region. The duration of pain was on average 31.2 months. The previous diagnosis was TMD for all patients. A final diagnosis and respective treatments were provided. For Eagle’s syndrome (1) the patient received a styloidectomy. Oropharyngeal tumor (1) was treated with chemotherapy. The patient with tension-type headache (TTH) (1) was treated with tricyclic antidepressants (TCAs). Trigeminal neuralgia (2) was treated with carbamazepine (1) and trigeminal percutaneous radiofrequency rhizotomy (1). Psychiatric disorders (2) received psychotherapy. Atypical odontalgia (1) was treated with TCAs. All, except for the patient with tumor, reported relief.

In summary, this group presented with different diagnosis for their chronic pain. Six patients previously received some form of oral surgery (dental extractions or periapical surgery), without improvement.

The final evaluation of this group was pain free in six, satisfactory improvement in one (TTH) and without improvement in one (oropharyngeal tumor).
Group III - Chronic myofascial pain – Ten patients (women) aging 42 to 70 years old (median 59±9) received a prior diagnosis of TMD. The average duration of pain was 6.8 years. Despite the diagnosis of TMD, pain control was unsatisfactory due to inadequate treatment and the presence of other causes. The final diagnoses were, respectively, TMD (5), TMD associated with fibromyalgia (2); TMD associated with traumatic trigeminal neuralgia (2); or TMD associate with trigeminal neuralgia (tic douloureux) (1). Treatment was performed accordingly: a) neuropathic pain; b) fibromyalgia or c) chronic myofascial pain.

Summarizing, in this group, the musculoskeletal facial pain was associated with trigeminal neuralgia, fibromyalgia or non-paroxysmal pain of the oral cavity. Four patients reported the beginning of the pain after facial surgery. Three patients reported oral surgery and one patient changed the dental prosthesis previously as the treatment of pain, without any improvement. The final evaluation of this group was: pain free in six and satisfactory improvement in four.

The Table 2 shows the general characteristics of the sample. The diagnosis of patients with organic pathology was achieved in accordance with the International Association for the Study of Pain (Table 3).

DISCUSSION

The original diagnosis was incorrect or incomplete in 80.7% of the cases. We find 11 different diagnoses among the 26 patients of this sample: pulpitis (7), leukemia (1), oropharyngeal tumor (1), atypical odontalgia (1), Eagle’s syndrome (1), trigeminal neuralgia (4), continuous neuralgia (1), TMD (9), fibromyalgia (2), conversion hysteria (2) and tension-type headache (1). All of the 26 patients were referred to our service with a prior suspect diagnosis of TMD. These patients failed to improve due to misdiagnosis.

The patients in this study were seen by an average of 4.6 dentists or physicians before arriving at our clinic. Only after appropriate treatment, adequate pain control was achieved in the majority of the patients remained pain free six-month follow up evaluation. The average of 4.88 health care, 70% of the patients saw a general dentist and 30% saw a physician, was found in another study about referral patterns for all types for facial pain.

Dental pain, trigeminal neuralgia and oncologic conditions may present with similar clinical symptoms. Tumors can provoke throbbing pain when compressing tissues, such as the patient with an oropharyngeal tumor who reported pain during the mouth opening movement. They cause neurological abnormalities too, as the left lipsparesthesia in the patient with leukemia. Trigeminal neuralgia is a sudden, usually unilateral, severe, brief, stabbing and recurrent pain in the distribution of one or more branches of the V cranial nerve; and seldom awakens the patient from sleep. On the other hand, pulpalgias can be triggered by cold or hot liquids, and can awaken the patient from sleep causing dental tenderness. Therefore, the use of specific diagnostic criteria is important to help in the differential diagnostic process. A simple example is seen in six of the seven patients that presented with pulpitis who described nocturnal episodes of pain. Three of these patients realized an increase in pain with an application of a external stimulus in their teeth (cold air). These pains were interrupted by the local anesthesia block.

This sample included two interesting cases of dental pain of nonodontogenic origin (acute leukemia and atypical odontalgia), and seven patients with craniofacial pain from odontogenic origin. This demonstrated that the location of pain is not always the same as its source. The atypical odontalgia, that is a neuropathic pain, is localized in the tooth or gingival, but the pain of pulpitis can vary greatly in its clinical presentation and intensity. Although trigeminal neuralgia (TN) has a well-defined diagnosis, it is often confused with other sources of facial pain with similar symptoms. This is due to the great variety of facial pain sources, the relative rarity of TN and the absence of the specific tests for its diagnosis. In this sample, we examined 5 patients with a diagnosis of TN and three of them were associated with TMD. Two patients presented with neoplasias (leukemia and nasopharyngeal tumor). This shows a necessity and importance of a differential diagnosis in orofacial pain, and demonstrated that pain may be an initial manifestation of a tumor. These cases also demonstrate the importance of the adjunctive testing such as CT and laboratory testing for diagnosis of pain. This also reinforces the fact that pain is a symptom that must be evaluated, especially when persistent, and despite treatment there is no significant improvement (5% of neuralgias can be symptomatic and secondary to tumor or neuronal diseases such as multiple sclero-
in this study idiopathic trigeminal neuralgia was the prevalent diagnosis.

This sample includes a patient with Eagle’s syndrome and another with tension-type headache. These patients also present with symptoms similar to TMD and we need the diagnostic criteria for these disorders. Another difficulty is in the differential diagnosis of orofacial pain that involves psychiatric disorders and simulates organic facial pain. This was the case of two patients with psychiatric disturbances who were eventually diagnosed with hysterical conversion disorder. Diagnosis of mental health disorders is a challenge that demands careful examination by specialists. The diagnosis of these cases was possible due to the fact that some pain conditions are uncommon (for example, pulpititis), leading to a misdiagnosis, iatrogenesis and chronicity of the pain. In this entire sample, the cause for persistent pain was perpetuated by an incorrect diagnosis and misdirected treatment. This demonstrates that a systematic evaluation, based on specific diagnostic criteria can help to clarify the diagnoses and formulate the treatment strategies for an appropriate therapeutic regimen. Finally, an interdisciplinary team is often necessary for the diagnosis and treatment of many facial painful conditions. The data presented in this study are consistent with data reported in other studies, indicating that while most orofacial pain is benign, there are cases where it may represent serious and even life threatening disease. This study also highlights the responsibility of the general practitioner, dentist or physician, to refer difficult patients for a more detailed and specialized evaluation.

REFERENCES