

Chronic postoperative orofacial pain. Case reports

Dor orofacial pós-operatória crônica. Relato de casos

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ABSTRACT

BACKGROUND AND OBJECTIVES: The presence of neoplasms, chronic and oral diseases may require surgical treatment for its resolution, although it may consequently cause chronic pain. Chronic postoperative orofacial pain remains even after tissue healing and its causes are not defined. Although neuropathic etiology is the most reported, it represents 30% of cases; the other 70% are still unclear and the main risk factors involved in the development of this chronic pain condition remains on discussion. The aim of the study was to report three clinical cases of different postoperative orofacial pain etiologies.

CASE REPORTS: **Case 1:** Female patient, 39-year-old, history of osteoblastoma exeresis in the mandibular body, presenting continuous postoperative shock pain, with intra and extraoral allodynia in the area. Diagnosis: post-traumatic trigeminal neuropathic pain. **Case 2:** Female patient, 30-year-old, diagnosed with refractory epilepsy and neurocysticercosis, complained of orofacial pain and bitemporal headache worse after craniotomy that treated the reported diseases. Diagnosis: post-craniotomy headache and orofacial pain. **Case 3:** Female patient, 49-year-old, with hereditary hemorrhagic telangiectasia, complained of pulsing in the alveolar ridge after extraction of three teeth, performed at different times. Diagnosis: Perception of orofacial pain secondary to systemic vascular disease.

CONCLUSION: Different surgical procedures, intra and extraoral, led to the development of orofacial postoperative pain in the reported cases, whose etiology is not only neuropathic. Prospective multidisciplinary studies are necessary in order to clarify the causes of orofacial postoperative pain.

Keywords: Craniotomy, Chronic pain, Facial pain, Osteoblastoma, Pain, Postoperative, Hereditary hemorrhagic telangiectasia.

RESUMO

JUSTIFICATIVA E OBJETIVOS: A presença de neoplasias, doenças crônicas e doenças bucais pode exigir tratamento cirúrgico para sua resolução, embora possa ocasionar dor crônica. A dor orofacial pós-operatória crônica permanece mesmo após a cicatrização tecidual e suas causas não estão claramente descritas. A etiologia neuropática, embora seja a mais relatada, representa 30% dos casos; os outros 70% não estão elucidados e ainda são discutidos quais os principais fatores de risco envolvidos no desenvolvimento desta condição de dor crônica. O objetivo deste estudo foi relatar três casos clínicos de indivíduos com diferentes etiologias de dor orofacial pós-operatória crônica.

RELATO DOS CASOS: **Caso 1:** Paciente do sexo feminino, 39 anos, com histórico de exérese de osteoblastoma em corpo mandibular, apresentou dor pós-operatória em choque, contínua, com alodínia intra e extraoral na área abordada. Diagnóstico: dor neuropática trigeminal pós-traumática. **Caso 2:** Paciente do sexo feminino, 30 anos, com diagnóstico de epilepsia refratária e neurocisticercose, queixou-se de dor orofacial e cefaleia bitemporal com piora após craniotomia para tratamento das doenças relatadas. Diagnóstico: cefaleia e dor orofacial pós-craniotomia. **Caso 3:** Paciente do sexo feminino, 49 anos, com telangiectasia hemorrágica hereditária, queixou-se de pulsar em rebordo alveolar após exodontia de três dentes, realizada em momentos distintos. Diagnóstico: percepção de dor orofacial secundária à doença vascular sistêmica.

CONCLUSÃO: Diferentes procedimentos cirúrgicos, intra e extraorais, levaram ao desenvolvimento da dor orofacial pós-operatória crônica nos casos relatados, de etiologia não apenas neuropática. Estudos prospectivos multidisciplinares serão necessários para esclarecer as causas desse quadro doloroso.

Descritores: Craniotomia, Dor crônica, Dor facial, Dor pós-operatória, Osteoblastoma, Telangiectasia hemorrágica hereditária.

INTRODUCTION

In 1998, a pioneering study identified surgeries and trauma as risk factors for the development of chronic pain in 40% of the evaluated patients¹.

Even today, There aren't many studies about the development of persistent postoperative pain after outpatient surgical procedures, although this condition, being chronic, causes great morbidity to patients and presents an important social impact². The moment of transition from acute to chronic pain remains under discussion and several risk factors have been described, classified in pre, intra and postoperative³ as genetic

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susceptibility, pain preexisting surgical intervention, psychological factors, intensity of acute postoperative pain, inadequate perioperative and postoperative analgesia, age and gender, risk of nerve injury and presence of chronic diseases⁴⁻⁹.

Acute postoperative pain (APOP) naturally occurs after surgical procedures and is associated with the inflammatory process¹⁰; its occurrence is protective¹¹ and is therefore beneficial for alerting the individual to a tissue injury¹². This pain tends to decrease according to the healing process⁴, varies strong intensity and is more intense immediately after surgery⁶.

On the other hand, the current definition of chronic postoperative pain, by the International Association for the Study of Pain (IASP), is: "pain that develops or increases in intensity after surgery and persists beyond the healing process for at least three months after surgery, excluding other causes such as pre-existing pain conditions, infections or neoplasms"; the pain must be located in the area that was handled during the surgery or projected in the territory of innervation of the injured nerve or referred to a dermatome after surgery/lesion in the deep somatic and visceral tissues¹³. This type of pain with strong intensity has an incidence of 2 to 10% of cases, but is not always correctly diagnosed, with 30% of these patients presenting neuropathic pain^{14,15}.

Regarding the craniofacial region, IASP suggests in the new International Classification of Diseases (ICD-11), which will come into force in 2022, the diagnostic criteria for post-traumatic headache/orofacial pain as a clinical manifestation after seven days of injury or trauma or seven days after the individual's gain of consciousness to report pain and that persists for more than three months¹⁶.

The prevalence of persistent pain after dental surgery is 5 to 13%¹³. There may be a variation of 0.5 to 5% for neuropathic origin after dental implant surgery¹⁷ and 0.5% after orthognathic surgery (mandibular bilateral sagittal osteotomy)¹⁸. Postoperative chronic orofacial pain (POCOFP) of neuropathic origin occurs in at least 5% of patients undergoing maxillofacial surgery¹⁹.

The most common dental surgeries that predispose individuals to sensitive abnormalities and persistent pain of neuropathic origin are dental extractions, mainly of third molars, implant surgeries and oral and maxillofacial surgeries, such as grafts, tumor removals and orthognathic surgeries^{6,17,18,20}. Thermal, tactile and painful sensory abnormalities are present in orofacial pains of etiologies different from neuropathic, such as musculoskeletal, as in temporomandibular dysfunctions (TMD), when compared to the control group; the hypothesis being the chronicity of pain as occurs in the phenomena of peripheral and central sensitization²¹. Despite the greater recognition of neuropathic etiology in the diagnosis of POCOF, neurovascular and musculoskeletal origins should also be considered, including in the orofacial region^{14,16,22,23}.

In neurosurgeries, such as craniotomies, the incidence of chronic postoperative pain can vary from 7 to 30% of cases; about 25% have severe pain¹³. In chronic post-craniotomies headaches, the prevalence varies from 24 to 29%²⁴⁻²⁶; about 29.3% of studied patients also presented chronic headache after suboccipital access²⁴. Studies with patients undergoing frontotemporal pterional craniotomies frequently present headache and/or chronic orofacial pain, which can be neuropathic or musculoskeletal²⁴⁻²⁶. A 30-day follow-up study showed that 80% of patients had bi-

lateral temporomandibular musculoskeletal pain and sensitive changes only on the operated side²⁷.

In order to prevent chronic postoperative pain, it's important to perform techniques that are as less traumatic as possible and to carefully dissect the surgical field, minimizing peripheral nerve injuries and local inflammations^{4,28}.

Preemptive analgesia in general medical surgeries is indicated for acting on acute neuroplastic responses and on the reversible central sensitization phenomenon, after intervention, and this measure may also work for neuropathic pain, however, the literature is still controversial as to its effectiveness^{3,4,28}.

In addition, perioperative analgesia and adequate post-surgical pain control are essential for the reduction of acute pain and, therefore, for the possibility of chronic pain^{3,6}.

The treatment of persistent post operative pain, given its complexity and challenge for the clinicians⁵, requires multidisciplinary teams and, in addition to that, the treatment for the global improvement of chronic post operative pain is not yet well defined²⁹. In this scenario, multimodal therapies are well indicated and include pain education, physical therapies and psychological interventions, such as cognitive-behavioral therapy and pharmacological treatment³, the treatment of neuropathic pain should be guided by the use of drugs of the first, second and third lines of scientific evidence^{30,31}.

On the other hand, treatment for chronic masticatory musculoskeletal pain (muscle TMD) secondary to craniofacial surgical procedures should also be considered. Moreover, the psychiatric/psychological follow-up of patients with POCOF is important, since many develop emotional and psychic alterations as a result of the pain condition, modulating it; the relation between pain and depression, for example, is very clear in the literature, but it is not yet known which one precedes the other²⁰.

The purpose of this study was to report three clinical cases of POCOF of individuals with distinct chronic diseases, submitted to different surgeries, indicating the variety of patients and surgical conditions which may be risk factors for its development.

CASE REPORTS

Case 1. POCOF after jaw osteoblastoma resection

Female patient, 39 years old, under clinical follow-up with the Orofacial Pain Team of the Dentistry Division of University of São Paulo School of Medicine Teaching Hospital (HC-FMUSP) Central Institute, complaining of continuous "electrical shock" mandibular pain of strong intensity, score 8 on the numeric pain scale (NPS), starting five months after tumor removal surgery. Lips movements triggered pain. On intraoral (IO) physical examination, the patient presented partial upper and lower dentition, with good oral hygiene condition, allodynia and hyperalgesia in central incisor teeth inferior to the left first premolar, in vestibular gum and ipsilateral jugal mucosa, limited mouth opening of 23mm, hypoesthesia in left lower lip, hyperalgesia in ipsilateral jaw base. Orofacial pain diagnosis: Post-traumatic trigeminal neuropathic pain³²/other trigeminal nerve disorders³³. Conduct: multidisciplinary treatment through neuropathic pain protocols, photobiomodulation sessions and mobilization of masticatory muscles with physical therapy.

Case 2: POCOPF after neurosurgery

Female patient, 30 years old, under clinical follow-up with the Orofacial Pain Team of the Dentistry Division of HC-FMUSP Central Institute, with a history of drug refractory epilepsy, in treatment since the age of two and monitored depressive disorder. Complaint of headache and chronic orofacial pain after craniotomy. The patient presented primary sleep bruxism, as well as bruxism secondary to epileptic seizures. At extraoral physical examination, she presented facial symmetry, intact skin, absent flogging, allodynia in right V1 territory, thermal and tactile hyperesthesia in right V2 and V3, hyperalgesia in left V1 and V3, preserved mandibular movements, without pain; moderate hyperalgesia on palpation in bilateral masseter muscle and in cervical muscles to the right radiating to the left; severe in temporal muscles; moderate to the right and severe to the left in posterior and lateral poles of temporomandibular joint (TMJ) in the static and dynamic exam.

On intraoral physical examination, the patient presented partial upper and lower dentition. Presented hyperalgesia to the horizontal and vertical percussion of the first premolar, second premolar, second molar and third molar to the lower left. On bidigital palpation, the myofascial trigger point (MTP) was located in the left masseter muscle with severe and moderate hyperalgesia on the contralateral side and MTP in the left temporal muscle. Orofacial pain diagnosis: Chronic post-craniotomy headache³⁴/chronic post-craniotomy orofacial pain/chronic post-traumatic headache and TMJ disorders³³. Conduct: physical therapy and occlusal stabilization splint, maintaining neurological treatment. Difficult development, refractory to treatments, including acupuncture and physiotherapy.

Case 3: Perception of chronic orofacial pain after exodontia in telangiectasia patient

Female patient, 49 years old, under clinical follow-up with the Orofacial Pain Team of the Dentistry Division of HC-FMUSP Central Institute with Rendu-Osler-Weber Syndrome (Hereditary Hemorrhagic Telangiectasia), Graves' disease, developing hyperthyroidism and mitral valve prolapse. The patient complained of upper lip and left face continuous, strong intensity (NPS=7), pulsing, not classifying it as pain, as well as in the region of exodontia performed at different times on the right upper canine teeth, right lower first premolar and left lower first premolar. Onset of symptoms two months after dental interventions. Improvement factors were jaw resting, cold compress and feed. Worsening factors were when the patient felt tachycardia and during physical efforts. The patient referred the same complaint of pulsing in the lower eyelid region and upper lip of the left side where the arteriovenous malformations (AVMs) were located and in distal upper and lower limbs, bilaterally. On intraoral physical examination, she presented partial upper and lower dentition, hyperalgesia on bidigital palpation in the left masseter muscle and hyperesthesia and hyperalgesia in the region of mentioned teeth. Orofacial pain diagnosis: perception of orofacial pain secondary to systemic vascular disease/other specified peripheral vascular diseases³³. Conduct: expectant/variable control; maintaining pulsing complaints of the same intensity in the described locations.

DISCUSSION

The reported cases show that chronic postoperative pain complaints range from classic neuropathic pain (Case 1), post-craniotomy pain, including orofacial pain (Case 2), to the pulsatile perception, not always easily distinguishable from throbbing pain, nevertheless, resulting from vascular disease (Case 3). These are unprecedented reports and their objective is to show the heterogeneity of the patients, especially in dentistry hospitals or oral medical services. It's important not to confuse the difficulty of treatment, as in cases 1 and 2, with situations that are very poorly defined or known, as in case 3. In the first case, for neuropathic pain, the immediate recognition in order to clarify the patient and to start treatment is not always simple. Case 2 also demonstrates the difficulty of a complex condition that requires a multiprofessional team for its management. Case 3 illustrates the difficulty of diagnosis and the thorough and multiprofessional study required to define the diagnosis and properly guide the patient.

In the third case specifically, Rendu-Osler-Weber Syndrome seems to have predisposed the patient to the pulsatile perception after teeth extraction³⁵. Although the patient presented some abnormalities sensitive on physical examination that may still point to neuropathic pain, the symptom in the area of the teeth extraction was equivalent to that of the AVMs locations (left lower eyelid and left upper lip), characterized as pulsatile and continuous. Sensitive abnormalities are not always strictly associated with the neuropathic component, and may correspond to areas of peripheral and central sensitization or the pathophysiology of the underlying disease²¹.

Cases like this require careful effort during anamnesis so as to understand the patient's complaint in order to allow a rational assessment. As it is a rare case, in which there was repeated complaint at each exodontia, the causal relationship between the surgical intervention and complaints seems to be the path to the diagnostic definition. Prospective studies with this disease and procedure may help to better understand this type of throbbing complaint.

Most of the studies found focus on the evaluation after general medical surgeries due to their higher prevalence, such as thoracotomies and mastectomies^{2,4}, and no prospective work has evaluated the prevalence of POCOPF after the various surgical modalities restricted to the dentistry field.

There is further discussion to be made around the subject of postoperative chronic pain and POCOPF. Although neuropathic pain is the most well-known, as already demonstrated by authors⁴, other conditions of persistent chronic pain remain an issue, as already reported by other studies^{22,24-26}, and may also be neurovascular and/or musculoskeletal, of nociceptive origin. It's important to emphasize that the presence of chronic diseases as in the three described cases may predispose the patient to the development of POCOPF⁷.

It's relevant that the dentist recognizes POCOPF as a disease and the fact that it's not necessarily associated with error in dental surgeries or procedures. The understanding of this problem reiterates the need of the dentist to understand that persistent pain after surgery generates a different diagnosis from the initial one, which must be correct in order to result in an adequate treatment

of post-operative sequelae and no longer intervene in an invasive manner in an attempt to minimize and solve the problem. Therefore, the persistence of pain must be thoroughly investigated in order to avoid unnecessary procedures that worsen the pain and compromise the result of the original surgery. Therefore, it's always important to differentiate POCOPF from infectious complications or those caused by other diseases¹³.

CONCLUSION

POCOPF is a reality, although it has only recently been recognized as a chronic pain scenario. It occurs in several types of oral or maxillofacial surgeries and even in other surrounding areas related to the dentist's performance. Although the neuropathic origin is the most well-known, there are other uncommon, less frequent or rare ones, and dentistry has the challenge of studying this scenario within the group of orofacial pains. Therefore, longitudinal and multidisciplinary studies are necessary to better understand this scenario of pain.

AUTHORS' CONTRIBUTIONS

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Writing – Preparation of the original, Writing – Review and Editing

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Writing – Review and Editing

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