IOSR Journal of Dental and Medical Sciences (IOSR-JDMS)
e-ISSN: 2279-0853, p-ISSN: 2279-0861.Volume 18, Issue 3 Ser. 7 (March. 2019), PP 71-75
www.iosrjournals.org

Paraesthesia Far From The Original Source of Pain in Occipital Neuralgia with Concomitant Craniomandibular Disorders.

Omar Franklin Molina[1] Marcus S. Peixoto[2] Maria A. Sobreiro[3] Zeila Coelho Santos[4]. Ed Wilson César[5] Jamil Elias Dib[6] Fábio Soares[7]

[1]*Department of Orofacial Pain, University of Gurupi, Gurupi-TO, Brazil*
[2]*Department of Orthodontics, University of Gurupi, Gurupi-TO, Brazil*
[3]*Department of Anatomy, University of Gurupi, Gurupi-TO, Brazil*
[4]*Department of Orthodontics, University of Gurupi, Gurupi-TO, Brazil*
[5]*Department of Operative Dentistry, University of Gurupi, Gurupi-TO, Brazil*
[6]*Department of Oral Surgery, University of Gurupi, Gurupi-TO, Brazil*
[7]*Department of Endodontics, University of Gurupi, Gurupi-TO, Brazil*
Corresponding Author: Omar Franklin Molina

Abstract
Introduction: *Occipital neuralgia is a complex neurophysiological disorder usually associated with local, referred pain and paraesthesia to distant areas from the original source of pain.*
Aim: *Assess frequency of paraesthesia, anatomic location and test the hypothesis that paraesthesia increases with longer duration of occipital neuralgia pain.*
Methods: *Clinical examination, palpation of joint and masticatory muscles, self-reported questionnaires and pain history were used to gather clinical and epidemiological data in 105 subjects presenting with occipital neuralgia and craniomandibular disorders. The experimental subgroup was compared with two control subgroups: Tension-type headache + Craniomandibular Disorders no occipital neuralgia and No Tension-type headache no occipial neuralgia and no CMDs.*
Outcome: *The frequency of paraesthesia was higher in the subgroup with occipital neuralgia and decreased in frequency from the ON + CMDs to the other two control subgroups (X-squared for independence p<0,0001, for trends p<0,0001). The frequencies of paraesthesia were 38,1%, 4,8% and 2,3% in the occipital neuralgia, Tension-type headache and no Crniomandibular disorders subgroups, respectively. Paraesthesia was observed in all subgroups with occipital neuralgia with different pain durations. There were no statistically and significant differences when the subgroups with different occipital neuralgia pain durations were compared regarding frequency of paraesthesia Its prevalence was higher in the "16 years or longer" duration of occipital neuralgia pain. Most common paraesthesia related sensations were numbness, tingling and burning most frequently reported in the face, tongue, inside the mouth and lips.*
Conclusions: *Although the frequency of paraesthesia was high in occipital neuralgia subjects, referred facial numbness was not reported in all subjects with signs and symptoms of occipital neuralgia. This investigation reinforces the notion that occipital is a neurophysiological disorder associated with both intense pain and paraesthesia in anatomic areas distant from the original source of pain.*

Keywords: *Occipital Neuralgia Craniomandibular Disorders Paraesthesia Pain Fifth Cranial Nerve.*

Date of Submission: 25-02-2019 Date of acceptance: 11-03-2019

I. Introduction

Craniomandibular disorders (CMDs) is a collective term used to describe a set of sign and symptoms affecting the masticatory muscles and temporomandibular joints (TMJs), usually of musculoskeletal origin presenting with specific signs and symptoms including a complaint of pain, joint noises, impaired jaw movements, tenderness to palpation of muscles and joints and headache[1]. CMDs sign and symptoms usually co-occur with neck disorders, facial pain and headache. Thus, TMJ, facial, headache and cervical pain occur concomitantly in a high percentage of individuals presenting with CMDs sign and symptoms[1].

Occipital neuralgia (ON) is defined by the International Headache Society as paroxysmal, shooting or stabbing pain occurring in the dermatomes of the greater and or lesser occipital nerve[2]. ON is a neuropathic pain condition due to nerve entrapment, whiplash injuries of the neck, tumor infiltration following neurosurgery, diabetes, inflammation of blood vessels and many other causes.[3] ON pain is usually described as paroxysms of stabbing, shooting pain reported in the occipital, sub-occipital and vertex regions[3],

commonly radiating to facial areas innervated by the fifth cranial nerve. Pain episodes may occur unilaterally or bilaterally. Other signs and symptoms including dysesthesia, paraesthesia, pain on neck movements, migraine - like symptoms, marked tenderness over the scalp and a pain generating zone have also been reported[3].

Paraesthesia is defined as a burning, prickling sensation or partial numbness caused by neural injury and may also be described as other associated sensations such as warmth, cold, aching, prickling, tingling, pins and needles, numbness, formication or itching[4]. Paraesthesia is an abnormal condition which causes an individual to feel a sensation of burning, numbness, tingling, itching or pricking frequently reported in the extremities, but it can be reported in other parts of the body as well and is associated with a multiplicity of etiologic factors[5]. Paraesthesia and numbness over the occipital scalp have been reported in many studies in patients presenting with ON[6]. However, investigations about referred paraesthesia to the territory innervated by the fifth cranial nerve are lacking in subgroups of patients presenting with ON and concomitant CMDs. Thus, this clinical investigation is aimed at:

1.Evaluate the frequency of paraesthesia in patients presenting ON and concomitant CMDs;
2.Assess the anatomic location of paraesthesia in the orofacial region in the same group of individuals.
3.Test the hypothesis that paraesthesia increases in frequency with longer duration of ON pain.

II. Material and Methods

Sample

All clinical charts of patients presenting with both CMDs and ON who sought diagnosis and treatment between 2010 and 2018 were retrieved from a database and examined retrospectively. This sample constituted the experimental ON subgroup (n=105). The first control group was constituted by 105 clinical charts from patients examined in the same period of time presenting with TTH and CMDs without ON. The second control subgroup was constituted by 44 subjects presenting with no CMDs, no ON and no TTH. Experimental and control subjects had been examined comprehensively in the period 2010-2018 using history of signs and symptoms, clinical examination of muscles and joints, muscle palpation, and self-reported questionnaires. During clinical assessment the principles of Helsinki declaration were followed: Patients were informed that their clinical evaluation and use of questionnaires had no absolute risk for their health, that any physical or psychological discomfort warranted the discontinuity of the evaluation, that accurate and comprehensive assessment was necessary in order to obtain accurate data and diagnosis before planning and treatment, that the principal examiner was scientifically experienced and qualified and that her/ his data would provide practical clinical benefits in future studies and treatments. Patients signed a formal consent allowing use of their clinical and demographic data for research purposes. Patients and controls were evaluated in the same period of time.

Thus, this retrospective investigation involved 105 subjects with ON and CMDs, 105 with CMDs without ON and 44 without CMDs and without ON.

Inclusion criteria for CMDs: Presence of TMJ noises, pain on palpation of the TMJs and/or masticatory muscles, difficulties to perform normal jaw movements, a complaint of muscle and/or joint pain, and seeking active treatment for CMDs.

Inclusion criteria for ON: A complaint of pain in the retroocular, vertex and occipital/sub-occipital areas, pain described as very intense, presence of nausea and vomiting, pain described as shooting or burning, pulsating, lancinating, intermittent and presenting with paraesthesia in the occipital area, reports of dizziness, ear stuffiness, and nasal constriction.

Inclusion criteria for TTHs and CMDs: A report of bilateral headache in the temporal, frontal and/or occipital/sub-occipital regions, described as dull, steady, pressing or compressing, moderate in intensity in both sides, a report of nausea (occasional), absence of vomiting and presence of trigger points in the cervical/and or neck areas directly associated with referred pain to the anterior part of the head.

Inclusion criteria for the second control group: Subjects in this group were those presenting a complaint in the masticatory system. However, they did not fulfill criteria for CMDs, for TTH or for ON.

Exclusion criteria: Subjects and controls presenting with severe psychiatric disorders, difficulties to respond properly to questionnaires, and presence of neuromuscular disorders including Parkinson´s disease, other epilepsy types, and speech and cognitive impairment were excluded from the comprehensive initial examination .

III. Statistical Analysis

Chi-squared for independence and trends, Kruskall-Wallis and Fisher´s exact tests were used in the current investigation.

IV. Results

There were 101 females=96,2% in the ON and CMDs subgroup, 101 females=96,2% in the TTH + CMDs subgroup and 31=70,5% females in the No CMDs subgroup: ON + CMDs versus TTH + CMDs

(Fisher´s exact test p=1,000); ON + CMDs versus No CMDs (Fisher´s exact test p<0,0001); TTH + CMDs versus No CMDs (Fisher´s exact test p<0,0001). Mean ages were about 39,5 (SD=9,6 range 18-81), 33,5 (SD=12,4, range 11-72), and 34,6 (SD=14,5, range 13-70) in the ON + CMDs, TTH + CMDs and No CMDs subgroups, respectively. Kruskal-Wallis´ statistics p<0,0001: ON + CMDs versus TTH + CMDs (p=0,001); ON + CMDs versus No CMDs (p<0,01); TTH + CMDs versus No CMDs (p>0,05). Mean age was lower in the No CMDs subgroup as it has been demonstrated that subjects in controls no CMDs subgroups are usually younger. See Table 1 for further details. The frequency of paraesthesia was much higher in the ON + CMDs subgroup (40/105=38,1%) as compared with the TTH + CMDs subgroup (5/105=4,8%) and with the No CMDs subgroup (1/44=2,3%). Fisher´s exact test ON + CMDs subgroup versus TTH + CMDs subgroup (p<0,0001); ON + CMDs versus No CMDs (p<0,0001); TTH + CMDs versus no CMDs (p=0,67). See Table 2 for additional details.

When ON + CMDs subjects with paraesthesia were allocated in certain subgroups characterized by different ON pain duration ranges, it was found that the frequencies of paraesthesia in these subgroups were about 9/22=40,9%; 8/27=29,6%; 6/20=30%; 5/15=33,3% and 12/21= 57,1%. Although the frequency of paraesthesia was higher in the last two categories with longer duration of ON pain, Chi-squared for independence and for trends tests yielded values of p=0,30 and p=0,23, respectively indicating that the subgroups were not independent and that there was no a trend to increased frequency of paraesthesia with longer duration of ON pain. See Table 3 for additional details. Finally, the most common sensations reported by patients with paraesthesia were numbness, tingling and burning. Anatomic sites reported by 40 ON/CMDs subjects with paraesthesia were the face (29/40=72,5%), tongue (9/40=22,5%), inside the mouth (6/40=15%), lips (5/40=12,5%), eyes/submandibular/TMJs (4/40=10%). See Table 4 for additional details.

V. Discussion

Higher frequency of facial paraesthesia was found in ON and CMDs subjects as compared with the two reference subgroups. Findings in the current investigation are contradicted by one study of case presentation[7] asserting that "the development of hemifacial sensory changes associated with referred trigeminal pain from ON is extremely rare". Because we observed a high prevalence of facial numbness in the current investigation, findings are supported by one investigation[8] in ON patients reporting a frequency of 41.1% of such a symptom. Referred facial paraesthesias or subjective numbness, especially in the cheek, is a greatly underreported manifestation of greater ON[9]. ON is considered by many researchers as a neuropathic disorder usually associated with nerve damage as a result of many types of traumatic events. The orofacial region may be the site of initial signs and symptoms of neurologic disorders which may manifest in the form of many pathological alterations including intermittent unilateral or bilateral facial numbness and/or pain, facial palsy and /or spasm[10]. Because a number of neurologic disorders, may cause facial paraesthesia, the outcome of the current study is in line with one investigation[11] indicating that paraesthesia in the form of facial numbness is a frequent symptom of neurological disorders.

Most common anatomic locations reported with paraesthesia were the **face, tongue, inside the mouth and lips. Thus, f**indings in the current investigation are supported by one case presentation[7] of ON involving the distribution of the left greater occipital nerve in which pain, paraesthesia and numbness progressively developed in the left hemiface of the patient. In one investigation[12] in which radiofrequency ablation was used in 23 patients presenting with cervicogenic pain with or without ON, researchers reported that the most common side effect of such a procedure was sub-occipital, ear and facial numbness which occurred in one out of 23 patients, implicating that facial numbness is related to ON and /or cervicogenic pain and that nerve damage may be a common cause. 22,5% of 40 subjects with CMDs, ON and concomitant facial paraesthesia demonstrated abnormal sensation in the tongue. Numbness in the tongue may be caused by compression of proprioceptive fibers coursing the tongue through the ansa hypoglossi, the cervical plexus and the second dorsal cervical root[13]. A traumatic event over the cervical area may cause multiple local and referred signs and symptoms including local pain, ON, numbness and tingling over the tongue[14] All subjects in the experimental group demonstrated signs and symptoms of both CMDs and ON using sound and widely accepted criteria. In the current study, paraesthesia was observed in a diversity of anatomic areas in the masticatory system including the face, maxilla and mandible, mucosa of the mouth, lips and tongue. It has been established that cervical involvement is an unrecognized source of orofacial pain including bilateral or unilateral neck and facial pain associated with numbness of the tongue, a symptom which may be brought on by a brisk, sudden axial rotation of the head[15]. Gallud and associates[11] evaluated a small sample (4 cases) of subjects presenting with multiple sclerosis and reported that all cases complained of numbness in the face. As a subset of cervicogenic headaches (CGH), ON can cause pain and paraesthesias to the posterior scalp, mandibular, periorbital, temporal and external ear regions[16]

In the current study, ON subjects were allocated to different subgroups based on the duration of their ON pain complaint. Although not statistically significant, the highest frequency of paraesthesia was observed

in the subgroup reporting the longest pain duration (16 years or longer). This may be so as frequency of paraesthesia distant to the original source of pain may be influenced by many factors including amount of nerve damage, pain duration, severer pain, increased local and central sensitization, concomitant pain or neurological disorders, other anatomical cervical disorders, posture and so on. Partially supporting this point of view, some researchers[7] defend the notion that increased sensitization of central nociceptive neurons in the cervical and trigeminal complex takes place in response to strong noxious input seen in some types of headaches. Elimination of the nociceptive input by decompressing a compressed anatomical structure may lead to complete relief of referred hemifacial pain and restoration of sensory loss[7].

Strong peripheral nociceptive input from second-order neurons in the trigeminocervical complex probably reduces pain threshold[17], enhances additional neural connections and produce enlargement of the receptive field, thus resulting in greater pain intensity, spreading of pain and altered sensations including paraesthesia to other anatomic areas. In trigeminal neuropathy, pain may dominate the clinical picture early on. However as the major cause of neuropathy progresses, pain becomes more chronic and neurons are destroyed, numbness and weakness usually appear[18]. The upper cervical spinal roots can contribute to sensory innervation of some cranial and cervical structures. Some occipital and sub-occipital structures such as blood vessels, dura mater, deep paraspinal neck muscles and adjacent muscle tendons and joints are recognized as sources of head and neck pain[7]. There is an anatomical overlap of trigeminal and cervical afferents in which many nociceptive afferents mainly those from C2 converge on the caudal segment of the fifth cranial nerve. Thus, sustained and strong nociceptive cervical input may converge in segments of the trigeminal complex causing not only sensitization but also nerve damage related neurophysiological changes including altered sensations carried from cervical areas to masticatory areas innervated by the trigeminal nerve. Such sensory changes are considered functional alterations in the central processing of noxious stimuli probably associated with strong and sustained nociceptive input and local injury.

VI. Conclusions

The current investigation is a study of a relatively large sample of subjects presenting with both ON and CMDs. In this investigation, ON subjects demonstrated the highest frequency of paraesthesia referred to areas innervated by the fifth cranial nerve. The highest frequency of such a neurological disorder was observed in subjects with ON with duration of 16 years or longer. The most common paraesthesia associated disorders were numbness, tingling and burning which were reported more frequently in the face, tongue, mucosa of the mouth and lips. Further studies in a larger sample of subjects with or without CMDs are necessary to map all types of paraesthesia associated disorders and the anatomic locations in which they are reported so as to further substantiate findings reported in the current study. To the best of our knowledge, this investigation is the first in the literature, reporting referred paraesthesia to the masticatory structures innervated by the fifth cranial nerve in a relatively large sample of ON subjects with concomitant CMDs.

Table 1: Social and demographic data in the experimental (n=105), Control 1= (TTH + CMDs=105) and Control 2= (No CMDs ==44) subgroups.

	SUBGROUPS					
	ON + CMDs n=105		TTH + CMDs n=105		NO CMDs No ON n=44	
GENRE	n	%	n	%	n	%
Females	101	96,2	101	96,2	31	70,5*
Males	4	3,8	4	3,8	13	29,5
Totals	105	100	105	100	44	100
Mean Age	39,5		33,5		34,6**	
SD	9,6		12,4		14,5	
Range	18-80		11-72		13-70	

*Fisher´s exact test: ON+CMDs versus TTHs + CMDs (p=1,000); ON+CMDs versus No CMDs (p<0,0001), TTH + CMDs versus No CMDs (p<0,0001).

**Kruskal-Wallis test p<0.0001: ON+CMDs versus TTH + CMDs (p<0,001); ON + CMDs versus No CMDs (p<0,01); TTH + CMDs versus No CMDs (p>0,05).

Table 2: Frequencies of paraesthesia in the experimental subgroup and in the control ones.

	SUBGROUPS					
	ON + CMDs n=105		TTH + CMDs n=105		No CMDs n=44	
FREQUENCY	n	%	n	%	n	%
Yes	40	38,1	5	4,8	1	2,3*
No	65	61,9	100	92,2	43	97,7

Totals	105	100		105	100		44	100	

*Fisher´s exact test ON + CMDs versus TTH + CMDs (p<0,0001); ON + CMDs versus No CMDs (p<0,0001); TTH + CMDs versus No CMDs (p=0,67).

Table 3: Frequencies of paraesthesia when the subgroups were organized hierarchically in increased order of ON pain duration (years).

	SUBGROUPS (ON PAIN DURATION: Years)									
	0-2		3-6		7-10		1-15		16 or longer	
Frequency	n	%	n	%	n	%	n	%	n	%
Yes	9	40,9	8	29,6	6	30	5	33,3	12	57,1*
No	13	59,1	19	70,4	14	70	10	66,7	9	42,9
Totals	22	100	27	100	20	100	15	100	21	100

*Chi-squared for independence p=0,30 For Trends p=0,23. There was a higher prevalence of paraesthesia in the subgroup "16 years of longer" with ON. However, there was no significant statistical difference when such frequency was compared with that in other subgroups.

Table 4: Types of referred paraesthesia and anatomic sites in the masticatory system reported by 40 subjects with ON, CMDs and paraesthesia

	Sensation	Frequency	%
	Numbness	30	75
	Tingling	6	15
	Burning	3	7,5
	Formicating	1	2,5
	Totals	40	100
Anatomic sites	Face	29	72,5
	Tongue	9	22,5
	Inside the mouth	6	15
	Lips	5	12,5
	Eyes	2	5
	Submandibular + TMJs	2	5

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Omar Franklin Molina. "Paraesthesia Far From The Original Source of Pain in Occipital Neuralgia with Concomitant Craniomandibular Disorders." OSR Journal of Dental and Medical Sciences (IOSR-JDMS), vol. 18, no. 3, 2019, pp 71-75.