

Occipital neuralgia as a true neuropathic pain: clinical and neurophysiological evidence

Neuralgia occipital como uma dor neuropática verdadeira: evidência clínica e neurofisiológica

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RESUMO

Objetivo. Avaliar a frequência de sintomas neuropáticos em indivíduos com neuralgia occipital, enxaqueca e dor de cabeça por tensão muscular e discutir os mecanismos da neuralgia occipital. **Método.** Critérios para essas dores de cabeça, distúrbios craniomandibulares e bruxismo, exame clínico, e questionários foram usados em 153 pacientes com distúrbios craniomandibulares e bruxismo. **Resultados.** As idades médias nos grupos foram 37,3±11,7 anos nos pacientes com neuralgia occipital, 36,5±11,8 anos nos pacientes com enxaqueca e 33,0±12,3 anos nos pacientes com dor de cabeça por tensão muscular. As frequências de dor tipo choque elétrico, dor em pontada, dormência, dor intensa, uma descrição de queimação, uma zona geradora da dor e dor intermitente foram de 54,3%, 77,1%, 34,3%, 100%, 68,6%, 100% e 57,1% respectivamente, nos pacientes com neuralgia occipital; 6,3%, 18,8%, 0%, 100%, 12,5%, 0% e 0%, respectivamente nos pacientes com enxaqueca, 0%, 17,6%, 0%, 18,6%, 0,9%, 0% e 0%, respectivamente nos pacientes com dor de cabeça por tensão muscular. A frequência da maioria dos sintomas neuropáticos esteve presente entre os pacientes com neuralgia mais do que entre os com dor de cabeça por tensão muscular. **Conclusões.** Os sintomas neuropáticos diferenciam neuralgia occipital de enxaqueca comum e de dor de cabeça por tensão muscular. A dor muito intensa é mais frequente nos pacientes com neuralgia occipital e enxaqueca, mas não se observa frequentemente nos indivíduos com dor de cabeça por tensão muscular.

Unitermos. Dor, Neuralgia, Cefaléia comum, Cefaléia Tensional.

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This investigation was carried out at UNIRG-School of Dentistry, Division of Occlusion and TMDs, Gurupi-TO, Brazil.

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ABSTRACT

Objective. Assess frequency of neuropathic symptoms in occipital neuralgia, migraine and tension-type headache, and discuss mechanism in occipital neuralgia. **Method.** Criteria for occipital neuralgia, migraine, tension-type headache, craniomandibular disorders, bruxing behavior, clinical examination, and questionnaires were used. **Results.** Mean ages are 37.3±11.7 years in occipital neuralgia patients, 36.5±11.8 years in migraine patients, and 33.0±12.3 years in tension-type headache patients. Frequencies of electric shock-like, stabbing or shooting pain, numbness, very intense pain, a burning description, a pain generating zone and intermittent descriptions were 54.3%, 77.1%, 34.3%, 100%, 68.6%, 100%, and 57.1%, respectively in occipital neuralgia patients, 6.3%, 18.8%, 0%, 100%, 12.5%, 0%, and 0%, respectively, in migraine patients; 0%, 17.6%, 0%, 18.6%, 0.9%, 0% and 0%, respectively, in tension-type headache patients. Comparing neuropathic symptoms between occipital neuralgia and migraine, and between occipital and tension-type headache, neuropathic symptoms were present almost exclusively in occipital neuralgia patients. **Conclusions.** Neuropathic symptoms differentiate occipital neuralgia from migraine and from tension-type headache. Very intense pain is more frequent in occipital neuralgia and migraine than in tension-type headache individuals.

Keywords. Pain, Neuralgia, Common Migraine, Tension-type headache, Migraine.

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INTRODUCTION

Occipital neuralgia is a specific pain disorder characterized by pain restricted to the sensory fields of the Greater or Lesser Occipital Nerves^{1,2}. Neuralgia is a painful paroxysmal disorder with sudden onset, rapid resolution, and very short duration (usually seconds or minutes)². Occipital neuralgia pain is localized in the occipital region due to the involvement of the occipital nerves³, and is described as a constant deep, burning pain with superimposed paroxysms of shooting and/or shock-like attacks, paresthesia and numbness over the occipital scalp².

Terms such as burning, aching, throbbing, and lancinating are used to describe occipital neuralgia, a pain type which occurs together with dizziness, nausea and photophobia⁴. Occipital neuralgia headache is also described as a unilateral or bilateral burning pain starting at the posterior base of the occiput, radiating behind the eye socket and laterally in the scalp⁴, sudden, intermittent, paroxysmic, pressing, throbbing, shooting, and sometimes lancinating pain which is reproduced locally by compressing manually the Greater Occipital Nerve⁵. Occipital neuralgia pain arises from trauma and/or entrapment of the occipital nerve within the neck of the scalp, or even from the C2-spinal root, C1-C2 or C2-C3 zygapophyseal joints². Neuropathic pains can be incapacitating, and are usually caused by a disorder or structural abnormalities affecting the peripheral nerves. Neuralgia, neuritis, neuromas and other neuropathies may cause pain in many anatomic areas and mimic odontogenic pain and craniomandibular disorders (CMDs)⁶.

C2 and C3 nerves innervate the back of the head, pierce the nuchal fascia and the base of the skull and thus, are prone to trauma from flexion, extension, injury and entrapment by spasm of the trapezius muscle⁷. A frequent mechanism in occipital neuralgia is sustained muscle activity in the cervical region, or entrapment of the C2 root or dorsal root ganglion by paravertebral ligamentous structures⁸. Greater Occipital Nerve compression and/or prolonged irritation by a nerve-vessel contact⁹, is a mechanism in occipital neuralgia and recommend the use of Greater Occipital Nerve decompression to relief intense pain. Occipital neuralgia is also associated with a myriad of symptoms in an anatomic area innervated by the trigeminal nerve, including the teeth. This is due to the con-

vergence of sensory information from the occipital and trigeminal nerve in the upper cervical chord¹⁰. Pain referral in occipital neuralgia arises from sensory connections between the main sensory nucleus of trigeminal nerve and the substantia gelatinosa of the cervical spinal cord, which is continuous with the inferior end of trigeminal nerve spinal tract nucleus¹¹.

The frequency and pathophysiology of some neuropathic symptoms in occipital neuralgia are not well documented neither clearly defined and some symptoms may occur in the trigeminal nerve territory. On the other hand, common migraine and even tension-type headache causes pain radiating to some trigeminal nerve areas including the maxilla and teeth. Occipital neuralgia may be confused with both cervicogenic headache and migraine¹². These controversies pose diagnostic and therapeutic problems for the clinician and call for further studies redefining these disorders through their signs and symptoms, for instance, there is a need to redefine occipital neuralgia as a neuropathic disorder. Consequently, the goal of this study is to compare the frequency of neuropathic symptoms in those CMD individuals with CMDs + occipital neuralgia, CMDs + migraine, and in those with CMDs + tension-type headache, and to discuss neuropathic symptoms mechanisms in occipital neuralgia.

METHOD

Sample

From a large sample of individuals (n=153) presenting with both signs and symptoms of CMDs and bruxing behavior referred consecutively over a period of ten years to UNIRG-School of Dentistry, Division of temporomandibular joint and orofacial pain, for diagnosis and treatment, all those presenting occipital neuralgia signs and symptoms (n=35), migraine (n=16) and tension-type headache (n=102), were selected and their charts were reviewed retrospectively so as to get data about the presence of neuropathic signs and symptoms. The investigation was approved by the Ethical Committee of the Dental School (005-2013) and only one expert in the field (OFM), reviewed patients' charts.

Procedures

Patients were classified as presenting CMDs if

they demonstrated at least two of the following signs, symptoms or behaviors characteristics of CMDs¹³: a complaint of facial and/or temporomandibular joint pain (TMJ), joint noises, muscle and/or TMJ tenderness on palpation, difficulties to perform normal jaw movements, seeking active treatment for a CMD complaint, and headache of TMJ and/or masticatory muscles origin. Exclusion criteria for CMDs: not seeking active treatment for a CMD complaint, presence of only one sign or symptom, for instance, joint noises, ear stuffiness, presence of neurologic and/or psychiatric disorders, inability to respond properly to some questionnaires and a report of dental pain described as very severe, throbbing, lancinating and electric-shock like which would be confused with neuralgic pain.

Occipital neuralgia: unilateral headache, continuous or paroxysmal, diminished sensation or disesthesia in the affected area, circumscribed tenderness over the Greater Occipital Nerve as it crosses the superior nuchal line, relief of acute attack by local anesthetic blocks, pain described as shooting and/or electric shock-like¹⁴. Exclusion criteria: Presence of other pain types, severe psychological and/or psychiatric disorders including dementia, difficulties to cooperate and give informed consent, inability to respond properly to questionnaires, and/or to cooperate during local anesthesia procedures.

Migraine: headache pain described as unilateral, moderate to severe in intensity, aggravated by physical activity, patient's report of nausea and vomiting, and sensitivity to light and sound¹⁵. Exclusion criteria: Other types of pain, pain described as always bilateral, unilateral pain described as mild, presence of severe psychological, behavioral and/or psychiatric disorders, inability to respond properly to questionnaires and/or unwillingness to sign a formal and written consent.

Tension-type headache: Headache described as being always bilateral, mild or moderate in intensity, pain occurring in the temporal, frontal and occasionally in the occipital regions and described as dull, constant, pressure and constriction, and presence of nausea more frequently than vomiting¹⁶. Exclusion criteria: presence of other headache types, unilateral headache, presence of severe psychological or psychiatric disorders, inability to respond properly to questionnaires and to cooperate and

sign a formal and written consent.

Patients were evaluated using comprehensive questionnaires with a number of descriptors for different headache types, establishing and obtaining a full description of the chief pain complaint including location, duration, frequency, intensity and quality, presence of some characteristics of neuropathic pain including the description of burning, stabbing, severe, electric shock-like and numbness, evaluating if the complaint was within the scope of CMDs, assessing muscle tenderness and trigger points by palpation, evaluation of jaw movements, use of diagnostic test for internal joint derangements and trigger points in the cervical region, assessing presence and severity of BB and oral jaw habits using appropriate questionnaires. Once we obtained clinical information and patients responded to self-reported questionnaires, they were classified as presenting CMDs and assigned to subgroups presenting occipital neuralgia, migraine, or tension-type headache.

Statistical analysis

Foi utilizado o Teste de Kolmogorov-Smirnov ou Shapiro-Wilk statistical method deemed to be appropriate in this investigation was the use of Fisher-exact test to assess differences in frequencies of neuropathic symptoms when comparing signs or symptoms in one pair of subgroups. Statistical significance was accepted if $p < 0.05$.

RESULTS

More females were represented in these subgroups of CMDs and bruxer patients presenting occipital neuralgia (85.7%), tension-type headache (91.2%), and migraine (100%). Mean ages were 37.3 years for occipital neuralgia, 33.0 for tension-type headache, and 36.5 for migraine (Table 1).

The frequencies of a electric shock-like pain description were 54.3% in occipital neuralgia, 6.3% in migraine, and 0% in tension-type headache groups. The frequencies of a shooting, jabbing or stabbing description were 77.1% in occipital neuralgia, 18.8% in migraine, and 17.6% in tension-type headache groups. Numbness was reported by 34.3% in occipital neuralgia, 0% in migraine, and 0% in tension-type headache groups, and very intense pain by 100% in occipital neuralgia,

Table 1

Demographic data in patients presenting occipital neuralgia, migraine, and tension-type headache patients.

CMDs + Bruxers	Occipital neuralgia (35)		Migraine (16)		Tension-type headache (102)	
	n	%	n	%	n	%
Females	30	85.7	16	100	93	91.2
Males	5	14.3	0	0.0	9	8.8
mean± standard deviation	37.3±11.7		36.5±12.3		33.0±11.8	

CMD = craniomandibular disorders.

100% in migraine, and 18.6% in tension-type headache patients. A burning description was reported in 68.6% in occipital neuralgia, 12.5% in migraine, and 0.9% in tension-type headache patients. A “pain generating area” was observed in 100% in occipital neuralgia, 0% in migraine, and 0% in tension-type headache group. A description of intermittent was reported by 57.1% in occipital neuralgia, 0% in migraine, and 0% in tension-type headache patients, as shown in Table 2.

The differences in frequencies of electric shock like pain was different between occipital neuralgia and migraine ($p<0.001$), occipital neuralgia and tension-type headache ($p<0.0001$), and non-significant between migraine and tension-type headache ($p=0.13$). There were differences in the frequencies of a description of shooting or stabbing were as follows: occipital neuralgia and migraine patients ($p<0.0001$), occipital neuralgia and tension-type headache patients ($p<0.0001$), migraine and tension-type headache patients ($p=1.0$). Numbness occurred exclusively in occipital neuralgia patients (34.3%), intense pain was a characteristic of both occipital neuralgia and migraine patients, and only 18.6% in tension-type headache patients described their pain as intense.

A burning description was reported more frequently by occipital neuralgia patients when compared to migraine ones (68.6% vs 12.5%, $p<0.0002$) and to tension-type headache patients (68.6% vs 0.9%, $p<0.0001$). A pain generating area was observed exclusively in occipital neuralgia patients as compared to migraine (100% vs 0%, $p<0.0001$) and to tension-type headache patients

Table 2

Frequency of neuropathic symptoms in occipital neuralgia, migraine, and tension-type headache patients

Headache types	Occipital neuralgia (35)		Migraine (16)		Tension-type headache (102)	
	n	%	n	%	n	%
Electric shock-like	19	54.3	1	6.3	0	0.0
Shooting/jabbing	27	77.1	3	18.8	18	17.6
Numbness	12	34.3	0	0.0	0	0.0
Very intense pain	35	100	16	100	19	18.6
Burning	24	68.6	2	12.5	1	0.9
Pain generating area	35	100	0	0.0	0	0.0
Intermittent	20	57.1	0	0.0	0	0.0

(100% vs 0%, $p<0.0001$). Intermittent was a description used exclusively by occipital neuralgia patients and the frequency was about 57.1% (Table 3).

DISCUSSION

Frequency of neuropathic symptoms in occipital neuralgia

One of the objectives of the current research was to assess the frequency of descriptors indicating that occipital neuralgia is typically a neuropathic pain disorder. Most patients in this study described their pain as electric shock-like, shooting, jabbing or stabbing, very intense, burning, and intermittent. Moreover, the presence of a “pain generating zone” was confirmed by gentle manual palpation in all occipital neuralgia patients. Only 34.3% occipital neuralgia patients described the presence of numbness posteriorly and anteriorly in the vertex area. Most descriptors used by the whole group on occipital neuralgia patients indicated that occipital neuralgia is a neuropathic pain disorder in character.

Electric shock-like

The frequency of a electric shock-like descriptor used by occipital neuralgia patients was much more frequent as compared to migraine and tension-type hea-

Table 3

Percentage of neuropathic symptoms comparing occipital neuralgia and migraine patients, occipital neuralgia and tension-type headache patients, and migraine and tension-type headache patients.

sign/symptom	Occipital neuralgia and migraine		Occipital neuralgia and tension-type headache		Migraine and tension-type headache	
Electric shock-like	54.3	6.3**	54.3	0.0**	6.3	0.0
Shooting/jabbing	77.1	18.8**	77.1	17.6**	18.8	17.6
Numbness	34.3	0.0**	34.3	0.0**	0.0	0.0
Intense pain	100	100	100	18.6**	100	18.6**
Burning	68.6	12.5**	68.6	0.9**	12.5	0.9*
Pain generating area	100	0.0**	100	0.0**	0.0	0.0
Intermittent	57.1	0.0**	57.1	0.0**	0.0	0.0

*p<0.05; **p<0.01.

dache patients. Thus, the outcome in the current study concurs with some investigations^{5,17,18} indicating that occipital neuralgia is typically described as electric shock-like. Occipital neuralgia is described as a paroxysmal or electric shock-like pain often superimposed over the constant pain of muscular origin² and/or as a continuous, electric shock-like and intermittent pain in nature⁴.

Shooting, jabbing, or stabbing

77.1% of the occipital neuralgia patients described pain as shooting, jabbing, or stabbing, thus, data in this investigation indicated that occipital neuralgia is typically described as shooting or stabbing and that such a description is probably the most common in occipital neuralgia patients^{5,19}. Shooting or electric shock-like pain is common descriptions used by occipital neuralgia patients².

Numbness

In the current study numbness was the descriptor used less frequently by patients. It may be that such a symptom is duration and intensity dependent in occipital neuralgia patients. This low prevalence of numbness is supported by one study, indicating that only 12 (33%) of the occipital neuralgia patients reported scalp pares-

thesia¹⁸. Further support for the outcome in the current study comes from one research²⁰ asserting that occipital neuralgia patients describe numbness and/or paresthesia anteriorly or posteriorly in the vertex region. Paresthesia and numbness over the occipital scalp are usually present in most occipital neuralgia patients².

Very intense pain

All occipital neuralgia patients in the current research reported the presence of very severe pain. The occipital neuralgia is described as very intense, extremely intense or even unbearable pain^{5,9,19,20}. Because of the paroxysmic nature of pain in occipital neuralgia, such a symptom is much more intense than pain in migraine and tension-type headache patients. Pain in tension-type headache patients is usually described as mild or moderate, however some tension-type headache patients particularly those chronic cases presenting with multiple trigger points in the neck, may present severe headache pain. In the current study, 19 (18.6%) tension-type headache patients reported the presence of severe pain. It may be that longer duration of pain, somatization traits, and contribution from multiple trigger points may cause severe pain in these tension-type headache patients. This assumption is highly speculative and merits further investigation.

Burning

The descriptor "burning" was used by 68.5% occipital neuralgia and 12.5% of migraine patients. Thus, this outcome concurs with other studies^{4,5} reporting that terms such as aching, burning, and lancinating are frequently used to describe occipital neuralgia pain. In theory, this is an unexpected finding, however, it might be the case that unrecognized occipital neuralgia in a migraine patient could result in the observation of symptoms typically observed in occipital neuralgia¹⁷.

Pain generating area

Because gentle and manual palpation demonstrated the presence of a pain generator area located caudally to the occipital protuberance, the outcome of this investigation is echoed by one study²⁰ asserting that possible

zones of Greater Occipital Nerve irritation and entrapment between the atlas and the axis, between the obliquus capitis inferior muscle and the semispinalis capitis and other areas, are observed in occipital neuralgia. The outcome reported in the current investigation is in line with one research²¹, indicating that occipital neuralgia is a refractory headache caused by irritation of Greater Occipital Nerve and or the Lesser Occipital Nerve causing “pain generating area” posteriorly in the head. Tenderness of the associated nerve along with the characteristic pain syndrome and relief of the symptoms with local anesthesia of the associated nerve, is useful in the diagnosis of occipital neuralgia.

Migraine patients in this study did not present a pain generating area, even though they may report scalp tenderness and allodynia¹⁷. Pain and tenderness in the back of the head is common in tension-type headache and migraine patients¹⁷, but such pain is more likely to be related to trigger points rather than to a pain generating area which is observed more commonly in occipital neuralgia individuals.

Intermittent

Many patients in the current study described their pain as electric shock-like and intermittent. This outcome has support in one study⁵ which described two clinical cases of occipital neuralgia and patients researchers evaluated described their pain as intermittent. In the current investigation, 57.1% occipital neuralgia patients described their pain as intermittent, thus the outcome of this study is in accordance with another investigation⁴, indicating that pain in occipital neuralgia is described as continuous and intermittent.

Neuropathic symptoms in occipital neuralgia compared to tension-type headache and Migraine

Neuropathic symptoms were very rarely observed in migraine and tension-type headache pain patients. Even though very severe pain is a characteristic of neuropathic pain, all migraine patients in the current investigation reported very severe pain whereas only 18.6% tension-type headache patients reported that their pain was very severe. This outcome is not an unexpected finding since migraine has been described in the literature as

severe unilateral headache²² and tension-type headache as a mild or moderate bilateral headache^{15,23}.

18.8% migraine and 17.6% tension-type headache patients described their headache pain as shooting or stabbing respectively. In the case of migraine, it may be that some migraine patients present some characteristics of occipital neuralgia, and/or alternatively that some patients present a combination of headaches. In many cases occipital neuralgia may produce migraine symptoms and various headache types may coexist in some patients¹⁷. Occipital neuralgia may also present in the context of migraine headaches with associated signs and symptoms²⁴.

Mechanism of neuropathic symptoms

Nerve contact, irritation, pressure and or entrapment are a common cause of neuropathic pains. The cause of cervicogenic pain is a nerve entrapment between the trapezius muscle, obliquus capitis inferior muscle or semispinalis capitis muscle²⁵. Greater Occipital Nerve compression by close proximity of the muscle tendon membrane and/or the swelling lymph node and/or the occipital artery in the peripheral course of the nerve may be an important mechanism causing pain and other occipital neuralgia symptoms⁹. It has been reported that entrapment of Greater Occipital Nerve causes occipital neuralgia and neurolysis of this nerve, particularly with regard to the trapezius aponeurosis is highly recommended²⁶. Moreover, the winding course of the greater occipital makes it more prone to compression at different anatomic points²⁶.

Trauma to the Greater Occipital Nerve may cause neurophysiological changes and the signs and symptoms are the result of an attempt of the nerve to regenerate itself. Tumors affecting the C2 and C3 nerve may also cause changes in the nerve membrane, and may account for the signs and symptoms characteristics of occipital neuralgia⁴. Prolonged contact, pressure or deformation of the nerve may cause superficial cutaneous sensitization which is currently considered an indicator of central sensitization and longer duration of occipital neuralgia headache¹⁷.

Nerve entrapment among the trapezius muscle, obliquus capitis inferior muscle or semispinalis capitis muscle may cause severe alterations in the nerve membra-

ne, being a common mechanism in occipital neuralgia²⁵. Local or systemic denervation, increased muscle activity in the cervical region and/or entrapment of the C2 root or dorsal root ganglion by paravertebral ligamentous structures, may cause neuropathic signs and symptoms⁸. The resection of the occipital nerve in 95 patients seen for chronic pain, demonstrated the presence of neuromata and scarring around the nerve, neural fibrosis and densely adherent occipital lymph nodes²⁷. These findings indicate that in many cases, structural local changes constitute a common mechanism in occipital neuralgia which ultimately results in neuropathic pain.

The pain sensitive structures may be stimulated by a variety of pathologic processes, including osteoarthritis and rheumatoid arthritis, temporal arterial inflammation, trauma, abnormal posterior, tumor, Paget's disease and entrapment between traumatized muscles¹⁸. Occipital neuralgia is described as a very severe pain which indicates cutaneous sensitization and increase pain duration¹⁷.

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