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CASE REPORT

Refractory cluster headache in a patient with bruxism and obstructive sleep apnea: a case report

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Abstract
Introduction This is a case report of a 39-year-old patient with a 14-year history of clinically refractory cluster headache (CH), also presenting obstructive sleep apnea (OSA) and complaining of tooth-grinding during sleep.
Discussion Treatment of OSA with an intra-oral device allowed an immediate reduction in frequency and intensity of CH events. Furthermore, CH attacks did not occur during the 12-month follow-up period.

Keywords Sleep apnea · Cluster headache · Orofacial pain · Oral appliance · Bruxism

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Introduction

The connection between inadequate sleep and headaches has been reported in medical literature, but its pathogenesis remains unclear [1]. Sleep-deprived normal subjects frequently develop bilateral frontal headaches described as pressure or squeezing [2]. However, it remains unclear as to how a cerebral disorder induced by sleep deprivation may activate nociceptive receptors of the meninges, blood vessels, and superficial structures [3].

Cluster headache (CH), chronic paroxistic hemicrania, and migraines are headaches frequently connected with changes in sleep patterns and specific sleep disorders [4].

CH are characterized by intense, paroxistic, and unilateral pain, associated with ipsilateral autonomic phenomena [5]. They may occur during sleep and are normally seasonal. Polisomnographic studies have revealed that sleep apnea in patients with CH was preceded by oxihemoglobin desaturation related to REM sleep [6].

Studies on the association between CH and obstructive sleep apnea (OSA) suggest that a reduction in mean oxygen saturation occurs, resulting in the hypothesis that recurrent hypoxemia may be the triggering factor for such events [7]. When both factors are associated, the treatment of sleep apnea may result in reduction or total remission of clusters [8]. For this reason, Nobre et al. have suggested that polysomnography (PSG) studies would be required to check the presence of OSA in patients with cluster headaches [9].

Sleep bruxism (SB) is a movement disorder characterized by tooth-grinding or clenching during sleep and associated with sleep arousals. Some subjects with SB also report facial pain, jaw discomfort, jaw lock, or temporal

headache upon awakening [10]. However, until now it is not clear what is the exact correlation between SB and chronic orofacial pain, including temporomandibular disorders (TMD) [11]. OSA has been found to be in association with sleep bruxism [12], but until now the relevance of these findings is not clear [13]. Some authors relate that masseter contraction classified as sleep bruxism was rare in patients with OSA; on the other hand, a mild elevation in masseter tone is frequently found during the termination of an apneic episode [13].

The diagnostic of OSA demands the need for treatment due to its significant health and economic consequences [14] and will be chosen according to the severity of the disease. When the OSA is mild, the oral appliance is considered the first choice [15]. Thus, the purpose of this study was to report a case of a patient with refractory CH that had a concomitant history of severe snoring and tooth-grinding during sleep that disturbed the sleep of his wife.

Case report

A 39-year-old male, a welder, complained of very intense, peri-orbital and maxillary crisis of pain (visual analogic scale (VAS)=10 (ranging from 0, absence of pain, to 10, the worst pain)) lasting about 2 h, along with autonomic signs (conjunctival redness, lacrimation, nasal congestion, rhinorrhea, and ptosis). Clusters took place during the day and while asleep, with an onset about 14 years ago. For the first 12 years, clusters took place once a year (normally in the month of December) and occurred daily, but later, their occurrence progressed to twice yearly. In the last 7 months, the patient presented 38 consecutive days with clusters that resulted in 17 trips to an emergency facility. During this period, he was excused from work for 7 days, and along with the CH attacks, he began presenting with less intense but daily and continuous facial pain. His wife reported frequently waking up at night because of his severe snoring and tooth-grinding. These complications lead them to look for dental treatment.

Patient assessment

The patient was submitted to a standardized protocol that consisted of an interview and systematic evaluation of cervical, cranial, facial, dental, and other oral structures according to the following specialized diagnostic instruments [10, 16]:

1. A standardized clinical orofacial evaluation to detail: (a) chief complaint; (b) general pain characteristics (location, intensity, quality, duration, pain relief, worsening); (c) patient's medical history with emphasis on headache and body pain complaints; (d) physical exam of the cervical, cranial, facial areas as well as intra-oral.
2. The Portuguese version of the Research Diagnostic Criteria for Temporomandibular Disorders (RDC/TMD) [17]

The following ancillary exams were carried out: hemogram, PSG, and a cranial computed tomography (CT). The hemogram and the CT have been used during the diagnosis of primary headache with the aim to exclude symptomatic diseases [5, 18]. In this case, both exams were normal.

The diagnosis of cluster headache was made according to The International Classification of Headache Disorders criteria [5].

IHS diagnostic criteria for cluster headache

3.1. Cluster headache

Diagnostic criteria:

A. At least five attacks fulfilling criteria B–D

B. Severe or very severe unilateral orbital, supraorbital, and/or temporal pain lasting 15–18 min if untreated

C. Headache is accompanied by at least one of the following:

1. Ipsilateral conjunctival injection and/or lacrimation
2. Ipsilateral nasal congestion and/or rhinorrhoea
3. Ipsilateral eyelid edema
4. Ipsilateral forehead and facial sweating
5. Ipsilateral miosis and/or ptosis
6. A sense of restlessness or agitation

D. Attacks have a frequency from one every other day to eight per day

E. Not attributed to another disorder

3.1.2. Chronic cluster headache

Description:

Cluster headache attacks occurring for more than 1 year without remission or with remissions lasting less than 1 month.

Diagnostic criteria:

A. Attacks fulfilling criteria A–E for 3.1 Cluster headache

B. Attacks recur over >1 year without remission periods or with remission periods lasting <1 month

The inclusionary criteria for TMD diagnosis include the presence of limited opening, tenderness of the masticatory muscles on palpation, and joint sounds during mandibular function, and the diagnosis of masticatory myalgia was made according to the Guidelines for Assessment, Diagnosis and Management of the American Academy of Orofacial Pain [19].

AAOP diagnostic criteria for masticatory myalgia

Local myalgia

Local myalgia is characterized by sore masticatory muscles with pain in the cheeks and/or temples on chewing, opening wide, and often on waking. It is usually bilateral and described as a stiff, sore, achy pain, spasm, or cramp.

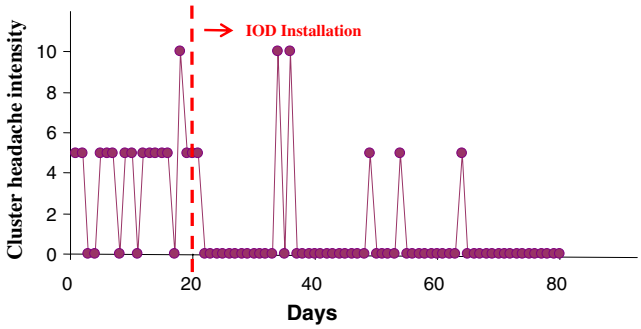

Fig. 1 Intensity of pain in the last 20 days before the onset of intra-oral device use and the first 60 days with the device. VAS 10 represents severe CH attacks, whereas VAS=5 represents moderate CH attacks and masticatory muscle pain

Diagnostic criteria

All of the following must be present:

1. Regional dull, aching pain during function of the affected muscle(s)
2. No or minimal pain at rest
3. Local muscle tenderness to palpation
4. Absence or trigger points and pain referral patterns

The following may accompany the preceding items:

- •Sensation of muscle stiffness
- •Sensation of muscle weakness
- •Sensation of muscle fatigue
- •Mouth opening may be decreased, but passive stretching of the elevator muscles will increase mouth opening by more than 4 mm (soft end-feel)

Physical examination disclosed severe dental wearing, intense masticatory muscle pain to palpation, and severe crepitation of the temporomandibular joint during mouth opening.

PSG disclosed OSA and rhythmic masticatory muscle activity (RMMA) do not relate to the apneas caused by apnea events.

Treatment

Since OSA was mild (AHI=11.4 events per hour), the patient was treated with an intra-oral device (IOD) for gradual mandibular advance used for every night's sleep, with daily activation of 0.25 mm for 2 months, which resulted in 9 mm advanced and approximately 70% protrusion rate.

Follow-up

Reductions in continuous spontaneous facial pain and CH attacks were noticed immediately after the second day of IOD use (Fig. 1). The patient was followed up for 12 months. He presented six episodic attacks in the first 45 days, but thereafter had neither facial pain nor cluster headaches.

Sixty days after the first PSG, we submitted the patient to another PSG (Table 1), a new clinical evaluation (the same baseline protocol), including masticatory muscle palpation, with the absence of muscle pain on palpation. At 6 months, we made a new clinical evaluation. After this, the pain complaint was evaluated through phone contact every 3 months until completing 12 months of follow-up.

Discussion

To our knowledge, this is the first report of cluster headache and OSA treated with an intra-oral device. The device was indicated because the apnea was mild, the dental wear was severe, and the patient presented discomfort of bilateral masseter muscles. Today, the effectiveness of oral appliances to control OSA cannot be denied [20], and studies found improvement in OSA even in devices without mandibular advancement [21], particularly when the severity of the problem and the presence of associated morbidities, such as obesity, are taken into consideration [22, 23]. Therefore, the use of the intra-oral device reduces apnea indexes and improves oxygen saturation [24]. Sixty days after the initial jaw advancement, improvements in several sleep parameters were observed (PSG), including a decrease in RMMA episodes (Table 1). The association between CH and OSA is not a new finding [1], and this clinical report illustrates the need for recognition of the several factors involved in this kind of headache, both to alleviate the patient's suffering and to improve the quality of life. Apart from its role in CH, OSA is considered a risk factor for a number of disorders, including SB, and early diagnosis is of fundamental importance [12].

Table 1 PSG results before (baseline) and 60 days after the use of a mandibular advancement device

	1st PSG (baseline)	2nd PSG (60days)
Total sleep time (min)	388	360
Sleep efficacy (%)	90.10	95
REM sleep latency (mins)	202	185
REM sleep (%)	18.10	21
Delta sleep (%)	13.80	14
Arousals	6.5/h	6.0/h
Minimal O_2 saturation (%)	83	92
Apnea/hypopnea index	11.4/h	4.3/h
Bruxism events	19	12
Clusters by event	3.68	3.08
Events per hour	2.94	2.0
Clusters per hour	10.8	6.16

In this case, we must understand that CH is one of the most severe types of pain described in literature and it has to be distinguished from masticatory muscle pain. The pattern and characteristics of these two types of facial pain are different. Certainly, the CH was the primary problem of this patient, but the presence of masticatory muscle pain raises the possibility of secondary or associated changes that may occur in patients with chronic or recurrent pain [25]. These changes might explain the presence of constant, mild to moderate facial pain in the late stage months. The presence of sleep bruxism may have contributed to the pain in the masticatory musculature. As for the orofacial pain complaint, mainly persistent, it is necessary to make a broad assessment of the patient, as there are innumerable sources of pain in this segment, and the very complexity of the trigeminal system contributes towards the spreading of the pain and difficulties of diagnosis.

In this case, interestingly, the patient had clinical symptoms of bruxism and was referred to our assessment for this reason. However, it is possible that the muscle component of facial pain is caused by the chronic pain itself and also by bruxism. So, things are different: the OSA is a recognized factor for worsening of CH, while bruxism is considered a risk factor for muscle pain [11], but not for CH. As the patient had constant pain attributed by the clinical evaluation to muscle pain, this may have a relationship with bruxism, but can also result from the central sensibilization that occurs in patients with chronic pain, persistent or refractory.

It is also known that OSA is considered a risk factor for SB [12]. Moreover, a previous study showed that devices for mandibular advancement reduce SB [24]. At 2 months, our patient presented a reduction of 37% (19 to 12 bruxism events) in RMMA episodes, but since this is only one case report, we cannot infer conclusions. However, the use of mandibular advancement may be a topic for further investigation.

So, we could infer that the use of the intra-oral appliance had two possible contributions: (a) improvement of the apnea and hypoapnea index with the consequent reduction of the CH crisis and (b) decrease of the sleep bruxism episodes and improvement of the masticatory muscle pain. However, we must not forget that the control of a severe pain, like CH, also contributes to reducing central sensitization and secondary effects, like muscle pain.

In conclusion, after the installation of the intra oral-device, CH attacks were controlled, sleep was normalized, and RMMA episodes were reduced. Although the association between CH and OSA is known in the literature, prospective studies are needed to evaluate the efficacy of intra-oral devices in these cases.

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