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CASE REPORT

An atypical survey in a cluster headache patient: A case report

Küme başağrılı bir hastada atipik bir araştırma: Bir olgu sunumu

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Summary

Cluster headache is a rare, severe headache associated with hypothalamic dysfunction or sleep cycles. It is classified in the primary headache group in The International Classification of Headache Disorders-3-2018 (ICHD-3-2018). In this case report, we present a 62-year-old male patient whose cluster headache showed a five times longer remission interval after dental implant treatment and ceased for more than two years following cardiac stent therapy.

Keywords: Cardiac stent; cluster headache; dental implant; remission.

Özet

Küme başağrısı, hipotalamik disfonksiyon veya uyku döngüleri ile ilişkili nadir görülen şiddetli bir başağrısıdır. Uluslararası Başağrısı Bozuklukları Sınıflaması'nda (ICHD-3-2018) primer başağrısı grubunda yer alır. Bu olgu sunumunda, ortalama yılda 2–3 küme atak periyodu geçiren, dental implant tedavisinden sonra küme başağrısı ataklar arası beş kat daha uzun remisyon gösteren ve kardiyak stent tedavisinden sonra iki yıldan fazla bir süredir atak geçirmeyen 62 yaşında bir erkek hastayı sunuyoruz.

Anahtar sözcükler: Dental implant; kardiyak stent; küme başağrısı; remisyon.

Introduction

Cluster headaches (CH) are rare but very severe, unilateral headaches occurring in the peri/supraorbital and temporal areas (trigeminal nerve, division 1). They appear in less than 0.4% of the population, with a male-to-female predilection of about 4–5 to 1.[1] Usually, they last between 15–180 minutes and are accompanied by trigeminal autonomic symptoms like lacrimation, tearing, conjunctival injection, nasal congestion, rhinorrhea, miosis, ptosis, facial or forehead sweating, and eyelid edema at the local area during a pain attack. Headache attacks typically occur once every other day to eight times a day. Each CH attack continues for weeks or months, usually at the same time of the day. We categorize it according to remission time, like more (episodic) or less (chronic) than 1 month.[2–4]

Another cause of headache in such patients is cardiac cephalgia. Although this type of headache is usually triggered by effort, it can be seen without any exertion activity. It can also be confused with migraine-type headache. Cardiac risk factors like hypertension, hyperlipidemia, cigarette smoking, diabetes, etc., often accompany this condition, and the most important sign is that cardiac cephalgia relieves after the treatment of myocardial ischemia/infarction.[5,6]

A third possibility in these patients is an overlap syndrome, characterized by the occurrence of two or more autoimmune diseases.[7] A temporal arteritis and trigeminal neuralgia overlap syndrome was identified by Gemici and Taşçı.[8]

In this report, we aim to present a patient with CH for more than 30 years, showing different remission intervals.

Case Report

A 62-year-old male patient was admitted to Aydın Adnan Menderes University, Faculty of Medicine, Department of Neurology with severe pain in the

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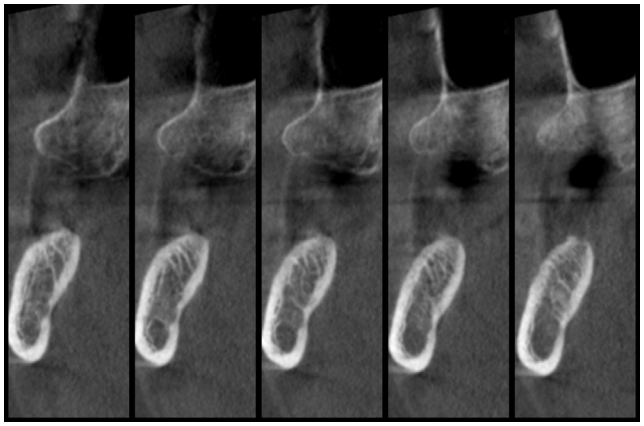

Figure 1. Cross-sectional images of the expansion region in the right posterior maxilla. No pathology was detected in the expansion region on maxillary Cone Beam Computed Tomography (CBCT) sections. Also, there was no pathology observed in the mandibular CBCT sections.

right peri/supraorbital area and temporal region. The patient had hypertension, diabetes mellitus, hyperlipidemia, and no significant family history. He had suffered from the disease for more than 30 years. The attacks continued between 90–120 minutes and repeated 3–4 times per day. Each cluster lasted 4–6 weeks, and he had more than a 1-month remission period (episodic type). His pain was rated 9–10 according to the numeric rating scale. He had verapamil 240–360 mg/daily, sodium valproate 1000 mg/daily, and melatonin 6 mg/daily for prophylactic treatment; and nasal oxygen with a mask by standing in a chair (10–12 L, 15–20 minutes), sumatriptan SC, greater occipital nerve (GON) and supraorbital nerve (SON) blockage for attack treatment during cluster periods. All these treatments had a partial effect, but indomethacin had no effect. We didn't try calcitonin gene-related peptide (CGRP) therapy, vagal nerve Gammacore stimulating therapy, and posterior hypothalamic stimulation therapy, which are known to be effective in CH.

The patient was admitted to a dental clinic when the pain did not sufficiently relieve with these treatments. The patient had root canal treatment for four teeth in the right posterior maxilla. When the pain still persisted, the patient had these four teeth extracted. Pain did not diminish during the two-year toothless period. A total of five dental implants were applied to the right posterior maxilla (3 pieces) and left posterior mandible (2 pieces) two years after these treatment approaches. Interestingly, the patient had no attacks for a year after

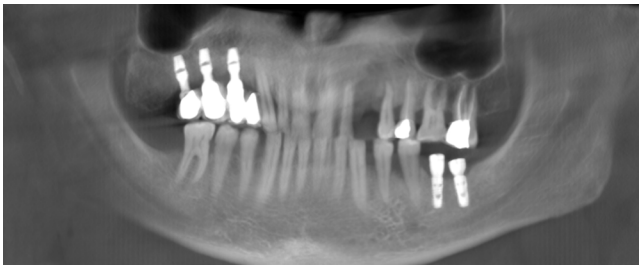

Figure 2. Panoramic image of the patient reconstructed from CBCT volumes. Mild alveolar bone loss was detected around the implants, but no obvious pathology was present.

the dental implant treatment. One year later, the patient applied to Aydın Adnan Menderes University, Faculty of Dentistry, Department of Oral and Maxillofacial Radiology for follow-up. No problem was observed in the patient's extraoral examination. No pain or tenderness was detected in the paranasal sinuses. When the temporomandibular joint (TMJ) was examined in open and closed positions, no problem was observed. Intraoral examination revealed bucco-palatal expansion in the right posterior maxilla. Cone beam computed tomography (CBCT) imaging was used to evaluate this expansion and available implants. No pathological finding was detected in the expansion region (Fig. 1). Mild alveolar bone loss was detected around the implants (Fig. 2). No pathology or abnormality was observed in the TMJ, nasal cavity, and maxillary sinuses entering the imaging field (FOV 16x10 cm).

Although there was a 4–6 week remission period before the implant treatment, there was a 1-year remission period after this treatment. One year later, his CH attacks came back again by the same modalities.

After the pain started again, he had an acute coronary syndrome, and coronary stent treatment was applied after coronary angiography. His CH attacks stopped after this surgical intervention for more than two years. He is taking esomeprazole, olmesartan/hydrochlorothiazide 20/10 mg, ticagrelor 90 mg, rosuvastatin 20 mg, acetylsalicylic acid 81 mg, and Humalog 50 every day.

After more than two years of follow-up, the patient still has no headache.

Written informed consent was obtained for the publication of the case report and accompanying images.

Discussion

The face and head and neck region possess complex anatomy and specialized sensory innervation. These anatomic and neural structures significantly impact pain patterns in the face, head, and neck region and are crucial in identifying the source of the pain.[9,10] These facial structures may play a role in generating pain in related areas, thereby making differential diagnosis confusing and complicated. In this complex framework, neural networks in the facial region may vary from person to person.[1,11,12] In the present case, it is noteworthy that the patient's pain was relieved for 1 year after dental implant treatment and ceased following coronary stent intervention and the new drug regimen related to diabetes, hypertension, hyperlipidemia, and antiplatelet regimen.

In our opinion, melatonin, which may play an important role in CH, had no role in this patient, because after the implant therapy, the level of melatonin did not change. We hypothesized that a neuropathic pain mechanism, such as a-beta fibers reinnervation or ephaptic transmission, might have a role in this patient's headache. However, he experienced allodynia during attacks and it had a cyclic pattern. We did not find any evidence supporting the teeth reflexology hypothesis. According to this hypothesis, some doctors believe that each tooth indicates diseases of a specific organ.[13] We could not find any tooth reflexology chart that suggests a tooth disease may play a role related to the hypothalamus. The trigeminocervical complex may have a role in this patient. One hypothesis is that dental implant treatment may reduce some inflammatory mediators and/or decrease vasodilation in this area. After the coronary stent treatment, the CH attack stopped; this may be related to the resolution of myocardial ischemia, which resolves intracranial homeostasis by decreasing inflammatory mediators and intracranial pressure.

It differs from other trigeminal autonomic headaches such as paroxysmal hemicrania (PH) and short-lasting neuralgiform headache attacks (SUNCT and SUNA) due to the pain duration (more than 60 minutes) and lack of response to indomethacin treatment.[2,4] These headaches are characterized by short-lasting intense headache attacks. The differences in attack duration and frequency, as well as the response to therapy, distinguish them from CH.[14]

It also differs from trigeminal neuralgia (TN); because pain attacks in TN last from seconds to 2 minutes. Additionally, there are trigger points in TN, but there were no trigger points in our patient. The first branch of the trigeminal nerve is rarely involved in TN but usually is in CH. On the other hand, autonomic findings like lacrimation, eyelid edema, and conjunctival injection, are only seen in a few cases of TN.[2,11,12]

Occipital neuralgia (ON) is a subgroup of neuralgia where the occipital nerves are injured or inflamed. Sensory deficit or dysesthesia accompany this severe headache, localized in the upper neck and back of the head.[15] There was no change in pain characterization during compression or distortion of the patient's neck, so it is not ON, which usually affects the posterior part of the head.[2,16]

It is different from TMJ headache, which is especially seen in women (our patient is a man), and it is known that CH is the most prevalent headache type in men.[11] There was no limited and asymmetric mandibular joint motion; we excluded this possibility after clinical and radiological examination of the patient.

We excluded thalamic pain and multiple sclerosis, which may cause relapsing and remitting facial pain, through normal cranial MRI results. Functional cranial MRI findings in cluster headache (CH) often reveal inferior hypothalamic grey matter activation ipsilateral to the headache side.[17] We only have normal cranial MRI findings; functional cranial MRI was not evaluated.

Persistent idiopathic facial pain (PIFP) often begins following minor surgical interventions or trauma. The pain is typically confined to a limited area on one side of the face, deep and poorly localized. Emotional stress can exacerbate the severity of the pain. The characteristics and location of PIFP can change over time.[18,19] Therefore, we excluded persistent idiopathic facial pain as there was no history of trauma or similar pain characteristics.

Head and neck pain is a common cause of referrals to physicians or dentists worldwide. The complexity and multidimensionality of pain in the head and neck region, coupled with the emotional, psychological, and social well-being of the patient, make the diagnosis challenging.[9,12] CH is one of the trigeminal autonomic headaches with distinct pain

characteristics.[3] While our case was consistent with CH in terms of pain characteristics, it also exhibited unusual remission intervals.

Conclusion

The headache in our idiopathic cluster headache (CH) case exhibited two atypical remission phases. First, after dental implant treatment, it resulted in a 1-year remission period, which is more than six times the usual remission duration. Secondly, although he did not meet the clinical criteria for cardiac cephalgia (except for cardiac risk factors), CH attacks ceased following coronary stent implementation for more than two years. We present this case to highlight that cardiac and/or dental diseases may play a role in the secondary pathophysiology of CH, despite the known involvement of ipsilateral hypothalamic and sleep dysfunction (neither REM nor NREM phase) in idiopathic CH.[2,4]

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References

1. Vgontzas A, Rizzoli PB. Nonmigraine headache and facial pain. Med Clin North Am 2019;103:235–50. [CrossRef]
2. Headache Classification Committee of the International Headache Society (IHS). The International Classification of Headache Disorders, 3rd edition. Cephalalgia 2018;38:1–211. [CrossRef]
3. Robbins MS, Starling AJ, Pringsheim TM, Becker WJ, Schwedt TJ. Treatment of cluster headache: The American Headache Society Evidence-Based Guidelines. Headache 2016;56:1093–106. [CrossRef]
4. Issı Z, Öztürk V. Cluster Headache. Türkiye Klinikleri J Neurol-Special Topics 2018;11:66–73.
5. Torres-Yaghi Y, Salerian J, Dougherty C. Cardiac cephalgia. Curr Pain Headache Rep 2015;19:14. [CrossRef]
6. Lipton RB, Lowenkopf T, Bajwa ZH, Leckie RS, Ribeiro S, Newman LC, et al. Cardiac cephalgia: A treatable form of exertional headache. Neurology 1997;49:813–6. [CrossRef]
7. Parente J, Mathurdas P, Wandschneider L, Aranha J, Siopa L. Overlap syndrome. Acta Med Port 2011;24(Suppl 3):719–24.
8. İnalkaç Gemici Y, Taşcı İ. Temporal arteritis and trigeminal neuralgia overlap syndrome: A case report. Agri 2019;31:206–8. [CrossRef]
9. Scrivani SJ, Spierings EL. Classification and differential diagnosis of oral and maxillofacial pain. Oral Maxillofac Surg Clin North Am 2016;28:233–46. [CrossRef]
10. Tassorelli C, Tramontano M, Berlangieri M, Schweiger V, D'Ippolito M, Palmerini V, et al. Assessing and treating primary headaches and cranio-facial pain in patients undergoing rehabilitation for neurological diseases. J Headache Pain 2017;18:99. [CrossRef]
11. Bernstein JA, Fox RW, Martin VT, Lockey RF. Headache and facial pain: Differential diagnosis and treatment. J Allergy Clin Immunol Pract 2013;1:242–51. [CrossRef]
12. Sessle BJ. Why are the diagnosis and management of orofacial pain so challenging? J Can Dent Assoc 2009;75:275–7.
13. Barret S. Meridian Tooth Charts Signify Poor Judgement. 2014. Available at: https://www.dentalwatch.org/questionable/toothchart.html. Accessed Nov 27, 2023.
14. Stubberud A, Tronvik E, Matharu M. Treatment of SUNCT/SUNA, Paroxysmal Hemicrania, and Hemicrania Continua: An Update Including Single-Arm Meta-analyses. Curr Treat Options Neurol 2020;22:42. [CrossRef]
15. Gözübatık-Çelik G, Buluş E, Uluduz D, Göksan B. Occipital neuralgia with atypic symptoms: A case report. Turk J Neurol 2014;20:135–7. [CrossRef]
16. Amasyalı SY, Akyol A. Other primary headache disorders. Türkiye Klinikleri J Neurol-Special Topics 2018;11:94–105.
17. Iacovelli E, Coppola G, Tinelli E, Pierelli F, Bianco F. Neuroimaging in cluster headache and other trigeminal autonomic cephalalgias. J Headache Pain 2012;13:11–20. [CrossRef]
18. Benoliel R, Gaul C. Persistent idiopathic facial pain. Cephalalgia 2017;37:680–91. [CrossRef]
19. Gerwin R. Chronic facial pain: Trigeminal neuralgia, persistent idiopathic facial pain, and myofascial pain syndrome-an evidence-based narrative review and etiological hypothesis. Int J Environ Res Public Health 2020;17:7012.