LETTER TO THE EDITOR

To The Editor

Cluster Headache with Temporomandibular Joint Pain

Keywords: Cluster headache, Headache, Temporomandibular joint, TMJ

A 31-year-old man presented to the headache clinic with a 10-year history of right-sided severe retro-orbital stabbing headache, right-eye tearing, and restlessness. Symptoms would last between 30 and 90 minutes, occurring once every other day to 5 times a day over the span of 5 weeks between January and February every year. His past medical history was significant for classical systemic Hodgkin lymphoma treated with 12 weeks of chemotherapy, followed by radiation therapy (30 Gy in 20 fractions) 5 years prior to presentation. His recent positron emission tomography scan demonstrated no evidence of active disease. Interictally, his neurological examination was unremarkable. His magnetic resonance imaging (MRI) head with sella sequence was normal. He was diagnosed with cluster headache (CH) and responded well to high flow (15 liters/minute) oxygen therapy. He did not tolerate subcutaneous sumatriptan and verapamil. He did not want to pursue other treatment options since he responded well to oxygen therapy. Aside from the right eye retro-orbital pain, tearing, and restlessness, he noticed a localized continuous excruciating throbbing pain at the ipsilateral temporomandibular joint (TMJ) during each attack. Between attacks, he denied any restriction, pain, discomfort, clicking, difficulty with chewing, or limitation of jaw movement. His TMJ pain would completely subside at the end of an attack. He was referred to a dental TMJ specialist who confirmed the pain location described by the patient was in the TMJ, but his jaw and TMJ physical examinations were unremarkable. Panoramic X-ray of the jaw demonstrated no signs of structural TMJ abnormalities. A computed tomography or MRI of the jaw and TMJ have been considered but were deferred by the patient.

Our patient demonstrated classical features of CH based on the characteristics and duration. Previous studies report that radiation of pain to the upper teeth, mandible, cheek, ear, and shoulder can be experienced in a CH attack but more commonly in chronic CH.[1] This case is unique as there is no literature on TMJ pain as a clinical manifestation during a CH attack. Our patient did not have features of trigeminal neuralgia, which can be seen in cluster-tic syndrome.[2,3]

Both central and peripheral systems play a role in CH, but the exact pathophysiology of CH remains unknown. The temporal and circadian aspects support a central theory, and the trigeminovascular-autonomic reflex has been widely accepted as part of the peripheral theory.[4] A few hypotheses could be postulated for the localized TMJ pain based on the peripheral theory.

Sympathetic neurons from the superior cervical ganglion reach the TMJ along the vessels and play a role in pain reception and monitoring of blood volume.[4] During a CH attack, patients may also experience miosis or ptosis related to disruption to the sympathetic neurons. Fibers from the superior cervical ganglion destined to innervate the eye are compromised by carotid dilatation as they traverse the carotid canal and may sustain neuropraxic injury.[4]

During a CH attack, there is an activation of the trigeminovascular-autonomic reflex with the release of calcitonin gene-related peptide (CGRP), and if CGRP is administered to a CH patient in an active disease phase, it triggers a CH attack. Increased levels of CGRP have been found in ipsilateral jugular vein blood during the active phase of CH.[5] This process is hypothesized to have a key role in the intense pain perception and in the associated distinctive vasodilation.[1,4] Similarly, previous report revealed higher levels of CGRP in TMJ synovial tissue from patients with TMJ pain compared with controls.[6]

A vasodilatory mechanism may play a role. During a CH attack, there is vasodilation in both the external and internal carotid arteries.[4] TMJ receives its arterial blood supply from branches of the external carotid artery, predominately the superficial temporal branch. Other branches of the external carotid artery, namely the deep auricular artery, anterior tympanic artery, ascending pharyngeal artery, and maxillary artery may also contribute to the arterial blood supply of the joint.[7]

Although CH classically involves the ophthalmic branch of the trigeminal nerve, it can also involve the maxillary and mandibular branches.[1,4] The mandibular nerve provides the main nerve supply for the TMJ. Additional innervation comes from the masseteric nerve and deep temporal nerves.[8] However, it is not clear why CH attack usually involves the ophthalmic branch of the trigeminal nerve and why some patients have different trigeminal involvement.

Free nerve endings, many of which act as nociceptors, innervate the bones, ligaments, and muscles of the TMJ. When bone tissue, ligaments, or muscles become irritated or injured, sensory signals are relayed along the trigeminal nerve.[7,8] Bruxism can potentially exacerbate TMJ pain setting off the trigeminovascular-autonomic reflex. However, this is less likely since our patient demonstrated no signs of bruxism from his oral and jaw examinations, but mild intermittent bruxism could not be ruled out completely.

Our case raises the possibility of TMJ pain as a clinical manifestation of CH. It would also be prudent to explore if TMJ pain can trigger a CH attack rather than simply a clinical manifestation. A large cohort of patients would be required to study the association of TMJ pain and CH.

Conflicts of interest

The authors have no conflicts of interest (financial or non-financial) to disclose relevant to this study.

Consent for publication

Informed consent was obtained from the patient.

Authors' contributions

TLHC examined and wrote the manuscript. DKK and WJB edited and reviewed the paper. All authors read and approved the final version of the manuscript.

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Tommy Lik Hang Chan
Division of Headache and Facial Pain, Department of Neurology & Neurological Sciences, Stanford University, Palo Alto, CA, USA

David Dongkyung Kim
Department of Clinical Neurological Sciences, Western University, London, ON, Canada

Werner J. Becker
Department of Clinical Neurosciences & Hotchkiss Brain Institute, Cumming School of Medicine, University of Calgary, Calgary, AB, Canada

Correspondence to: Tommy L.H. Chan, MBBS, Division of Headache and Facial Pain, Department of Neurology & Neurological Sciences, Stanford University, Palo Alto, CA, USA. Email: tlhchan@stanford.edu

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