

# Cluster Headache-like Facial Pain following Dental Extraction: A Case Report

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A 50-year-old female patient with severe unilateral pain in the right eye, head, and face accompanied by lacrimation and drooping of the right eye and rhinorrhea from the right nose, which developed immediately after extraction of the maxillary right first and second molars, was successfully treated with oral administration of sumatriptan and prednisolone, or verapamil. Although the clinical characteristics are similar to those reported in cluster headache except the temporal feature, the probable cluster headache, the hemicrania continua and the acute migraine headache should be included in the list of differential diagnoses.

**Key Words:** Cluster headache; Facial pain; Migraine; Sumatriptan; Tooth extraction

## INTRODUCTION

Cluster headache is a stereotyped primary pain syndrome characterized by strictly unilateral severe pain, localized in or around the eye and accompanied by ipsilateral parasympathetic autonomic symptoms such as lacrimation, conjunctival injection, nasal congestion or rhinorrhea. Cluster headache usually affects patients aged 20-60 years and has a circadian or circannual periodicity.<sup>1)</sup> The prevalence is less than 1% of the total population, and men are afflicted three times more often than women.<sup>2)</sup>

The pathogenesis of cluster headache is complex and remains incompletely understood. The most widely accepted theory is that primary cluster headache is characterized by hypothalamic activation with secondary activation of the trigeminal autonomic reflex, probably via a trigeminal hypothalamic pathway.<sup>3,4)</sup>

Although cluster headache is regarded as a primary headache, there are many case reports in the literature suspecting symptomatic or secondary cluster headache or cluster headache-like headache. Some case studies describe cluster headache with ipsilateral arteriovenous malformations.<sup>5,6)</sup>

Some patients with cluster headache revealed intracranial neoplasms.<sup>7-10)</sup> Several reports have described the association of trigeminal autonomic cephalalgia-like headache with intracranial lesions,<sup>11)</sup> multiple sclerosis,<sup>12)</sup> or posttraumatic head injury.<sup>13)</sup>

In this report, we present a patient who developed cluster headache-like facial pain after the extraction of ipsilateral molars, which was responsive to combination therapy of sumatriptan and prednisolone, or verapamil.

## CASE REPORT

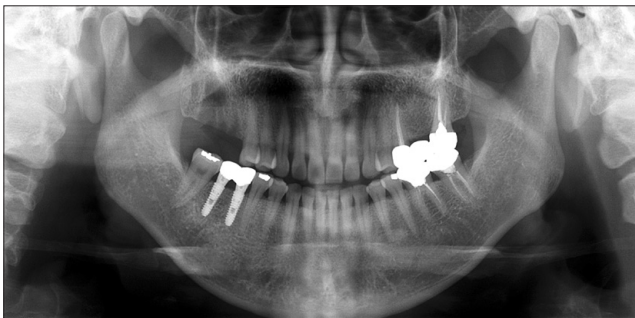
A 50-year-old female patient was sent from a local dental clinic to the Department of Oral Medicine, Kyungpook National University Hospital. The patient has had severe excruciating pain in the right eye and the right ear and severe pricking, stabbing, or aching pain in the right side of the face, the head, and the neck. The pain was quite continuous, not remitting all through the day, so the patient appeared anxious, restless, and agitated during initial examination. She also complained that the pain was accompanied by lacrimation and drooping eyelid of the right eye,

sense of pungent smell and rhinorrhea from the right nose. The condition started 2 months ago, immediately after the extraction of right maxillary first and second molars due to periodontal problem.

Before visiting our department, she had consulted a variety of dentists/physicians including general dental practitioners, an oral surgeon, an otolaryngologist, and a neurosurgeon. She had been undergone a number of examination including brain magnetic resonance imaging (MRI) and facial computed tomography. However, the neurosurgeon reported no structural abnormalities from the brain MRI. Maxillary sinusitis was also ruled out by the otolaryngologist. From the previous physicians or dentists, she had received a lot of medical treatment including antibiotics, analgesics, and gabapentin, but she didn't get any relief of pain from the medical therapy. On the contrary, she had suffered from severe gastric pain and symptoms of liver dysfunction resulting from heavy use of non-steroidal anti-inflammatory drugs (NSAIDs) or other drugs for pain control.

On clinical examination, the extraction site healed well with normal mucosa. The panoramic radiograph showed no pathologic findings in the jaw (Fig. 1).

Considering the location and severity of the pain and no responsiveness to previous medications including NSAIDs and anticonvulsants, we tentatively diagnosed the condition as cluster headache-like facial pain. When sumatriptan succinate 100 mg and prednisolone 20 mg were administered with divided dose in a day as an initial trial treatment for first 3 days, the pain decreased significantly and subsided 80%. Then the pain completely disappeared after daily administration of sumatriptan succinate 150 mg



**Fig. 1.** Panoramic radiograph of the patient. The extraction sockets for the maxillary right first and second molar are seen, but there is no pathologic findings related to the extraction.

and prednisolone 10 mg for next 1 week. After 2 months of the combination therapy, there was no recurrence of the pain. Then verapamil began to be administered with 40 mg, three times a day as a long-term preventive remedy in combination of sumatriptan succinate. The patient was free from the pain with administration of sumatriptan succinate 25 mg and verapamil 40 mg, twice a day.

## DISCUSSION

According to "The International Classification of Headache Disorders, 3rd edition (beta version)" published by the International Headache Society (IHS),<sup>14)</sup> the trigeminal autonomic cephalalgias (TACs) are a group of primary headache disorders that share the clinical features of headache, which is usually lateralized, and often prominent cranial parasympathetic autonomic features, which are again lateralized and ipsilateral to the headache. The TACs include cluster headache, paroxysmal hemicrania, short-lasting unilateral neuralgiform headache attacks with conjunctival injection and tearing (SUNCT), short-lasting unilateral neuralgiform headache attacks with cranial autonomic symptoms (SUNA), and hemicrania continua.

There is much overlap in the diagnostic features of individual TACs. In contrast, treatment response is relatively specific and it aids in establishing a definitive diagnosis. Cluster headache is very stereotyped in its presentation and fairly easy to diagnose with an in-depth headache history.

The diagnostic criteria for cluster headache published by the IHS in 2013 are as follows<sup>14)</sup>:

- A. At least five attacks fulfilling criteria B-D
- B. Severe or very severe unilateral orbital, supraorbital and/or temporal pain lasting 15-180 minutes (when untreated)
- C. Either or both of the following:
  1. At least one of the following symptoms or signs, ipsilateral to the headache:
    - a) Conjunctival injection and/or lacrimation
    - b) Nasal congestion and/or rhinorrhea
    - c) Eyelid edema
    - d) Forehead and facial sweating
    - e) Forehead and facial flushing
    - f) Sensation of fullness in the ear

g) Miosis and/or ptosis

2. A sense of restlessness or agitation

D. Attacks have a frequency between one every other day and eight per day for more than half of the time when the disorder is active.

E. Not better accounted for by another diagnosis described in the 3rd edition of "The International Classification of Headache Disorders (ICHD-3)."

Our patient showed unilateral severe, excruciating pain in the right eye along with severe pain in the adjacent face and forehead, which was accompanied by the ipsilateral parasympathetic autonomic symptoms such as lacrimation and rhinorrhea as well as the pain was responsive to sumatriptan instead of NSAIDs or gabapentin. Therefore, it is reasonable to assume that the patient should have cluster headache, based on the above criteria.

However, we couldn't make the confirmed diagnosis because the temporal feature of the pain bouts was pretty different from those described in the diagnostic criteria of cluster headache and the acute nature of the condition. In this case, the pain attack was so long in duration that the patient might feel it continuous rather than intermittent, and there was no remission period during the day. Therefore, she couldn't tell how often the pain attack occurred in a day and how long the pain lasted. She exclaimed she suffered from the pain all through the day.

This case fulfills all diagnostic criteria for cluster headache except longer duration of pain attacks. At this point, we don't know whether this case is a new type of trigeminal autonomic cephalalgia or it is just a variant of cluster headache. Further investigation and more clinical data are required to answer it. We think that the 'probable cluster' can be a possible diagnosis for this case. According to the IHS classification,<sup>14)</sup> the probable cluster headache has diagnostic criteria similar to that of cluster headache except permitting missing of any one of the criteria for cluster headache. In other words, it requires fulfilling all but one of criteria A-D for cluster headache.

Hemicrania continua is a condition to be considered as another possible diagnosis for this case, because the clinical features are very similar to those appeared in this case. Hemicrania continua is characterized by a continuous, unilateral headache that varies in intensity, waxing and

waning without disappearing completely. In addition, there are usually mild autonomic symptoms such as lacrimation, conjunctival injection, nasal symptoms, and ptosis/miosis.<sup>15)</sup> However, we were not able to choose it as a diagnosis for the case, because the case didn't respond to NSAID and the responsiveness to indomethacin was uncertain from reviewing of the patient's history. On the contrary, the case responded well to sumatriptan and verapamil.

Acute migraine headache also should be included in the list of differential diagnoses for this patient, especially because the oral administration of sumatriptan was effective for pain control in this case and migraine is also characterized by recurrent moderate to severe unilateral headaches often in association with a number of autonomic nervous system symptoms.<sup>14)</sup>

It is known that sumatriptan shows effectiveness in the acute treatment of cluster headache attack only when it is administered parenterally, not orally.<sup>16)</sup> However, in this case, the severe unilateral headache was slowly relieved by oral administration of sumatriptan. The pain subsided 80% at 3 days after sumatriptan therapy, and completely subsided after next one week. The pain attack didn't recur during the preventive administration of sumatriptan and prednisolone or verapamil. More information is needed to make it clear whether oral administration of sumatriptan is not effective at all for abortion of cluster headache attack or it can result in slow relief of the attack. To the best of our knowledge, this is the first report describing oral administration of sumatriptan for control of cluster headache-like facial pain which developed after dental extraction.

Regarding to relevance in cluster headache and dental practice, we found 2 case reports describing an association of cluster headache and dental extraction<sup>17)</sup> or dental treatment<sup>18)</sup> and one case report dealing with cluster headache resulting from an impacted upper third molar.<sup>19)</sup>

Sörös et al.<sup>17)</sup> reported two patients who developed cluster headache 2 weeks after extraction of ipsilateral molar tooth. They used oxygen to terminate the cluster headache attacks and verapamil for prophylactic treatment. Although the correlation between cluster headache and dental extractions seems to be mere coincidence, they strongly suggested the diagnosis of secondary cluster headache because of the following reasons: (a) the headache disorder exactly meets the

IHS criteria for cluster headache, (b) the headache attacks are responsive to standard acute and prophylactic treatment of cluster headache, (c) cluster headache attacks are ipsilateral to the extraction site, and (d) a close temporal relation between the development of the structural lesion and the onset of cluster headache exists.

On the other hand, Shoji<sup>18)</sup> stated that the orbital pain may refer to maxillary or mandibular areas, and the teeth in those areas and it is not uncommon for patients with cluster headache to consult a dentist, and to be initially misdiagnosed and receive unnecessary treatment. Furthermore, he didn't agree with the possibility of development of the secondary cluster headache following dental treatment, proposed by Sörös et al.<sup>17)</sup>

At present, while there is considerable clinical evidence for an association between brain or head lesions and the development of cluster headache,<sup>11,20)</sup> the crucial question how a head injury or a structural brain disease could contribute to the development of cluster headache is still undetermined. In addition, it is also unclear whether cluster headache-like facial pain may result from dental extraction or both events are just coincidental. Further investigation and more clinical data are required to explain this issue.

In summary, a 50-year-old female patient with severe unilateral pain in the right eye, head, and face accompanied by lacrimation and drooping of the right eye and rhinorrhea from the right nose, which developed immediately after extraction of the maxillary right molars, was successfully treated with the administration of sumatriptan and prednisolone or verapamil. This case report underlines the importance of proper diagnosis of the unusual facial pain, which may develop in association with dental treatment.

## CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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