

Cluster Headache Associated with Secondary Unilateral Blepharospasm: A Case Report and Review of the Literature

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Abstract

Purpose: Cluster headache is one of the most serious types of headache that is accompanied by autonomic parasympathetic symptoms. Its association with hemifacial spasm in the same side had been rarely reported. The aim of this report is describing a case with this association and treatment strategies.

Case Report: Here we report a 37-year-old female with cluster headache associated with secondary unilateral blepharospasm that was successfully treated with combination therapy including botulinum toxin injection.

Conclusion: Hemifacial spasm associated with cluster headache needs special attention and can be treated successfully.

Keywords: Cluster Headache; Disport; Secondary Unilateral Blepharospasm

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INTRODUCTION

According to the International Headache Society, cluster headache is characterized by episodes of unilateral pain in the supraorbital and temporal regions and occurs in the territory supplied by the ophthalmic division of the trigeminal nerve. If not treated, the pain typically lasts for 15 to 180 minutes. Facial autonomic symptoms include ipsilateral tearing, conjunctival injection, rhinorrhea, and nasal congestion.^[1] Cluster headache belongs to a group of disorders characterized by unilateral pain in areas supplied by the trigeminal nerve. The pain

occurs in association with prominent ipsilateral cranial autonomic symptoms, which are called trigeminal autonomic cephalgias (TACs).^[2] The hallmark of cluster headache is the circadian or even circannual periodicity, and studies have proven hypothalamus activity during attacks.^[3] Most patients experience cluster headache episodes that last days to months, thereafter going into remission. However, in 10% of the patients, the headache is chronic, without any relief between episodes.

Abortive therapy for the acute attack phase includes oxygen therapy, sumatriptan injections, sumatriptan nasal spray, other forms of triptans, and conventional preventive medications such as verapamil, lithium, cortisone, sodium valproate, and indomethacin. Surgical methods, particularly the destruction of the trigeminal ganglion, have also been used.^[4]

Botulinum toxin A may reduce the sensitization of the peripheral trigeminal afferent nerve, which results

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in the reduction of the nociceptive input and subsequent attenuation of central pain sensitization.^[5] Retrograde transmission of botulinum toxin A may modulate the release of neurotransmitters such as substance P^[6] or CGRP^[7] in the trigeminal nerve terminals. Therefore, botulinum toxin A injection to the pericranial muscles is used for maintenance therapy in headache syndrome as well as cluster headache.^[8-10]

As we reviewed the literature, we found only one report on hemifacial spasm associated with cluster headache^[11] and another one on bilateral blepharospasm in a case of TAC-like headache.^[12]

Here, we describe the case of a woman with a history of cluster headache and ipsilateral secondary blepharospasm who did not respond to the above-mentioned therapy. However, her symptoms subsided on administering combination therapy.

CASE REPORT

A 37-year-old woman with a complaint of left-sided headache and ipsilateral blepharospasm was referred to our oculoplastic clinic. The symptoms began 10 years ago as severe left-sided headache that gradually involved the entire head; the headache was not pulsatile but was sometimes associated with nausea. Primarily, the headache was episodic; however, over time, it became constant and was accompanied by mild depression and anxiety symptoms. At the time of headache, left-sided unilateral blepharospasm, ipsilateral photophobia, tearing, and rhinorrhea were also present [Figure 1a]. She often experienced headache at night and at waking time. The recurrence rate was very severe, and she was free from headaches only a few days a month. When the headache remitted, the blepharospasm also improved. In the last 10 years, the patient had received five Dysport injections in the left periorbital area, which induced

provisional remission of blepharospasm but had no effect on the headache. The patient had also systemic hypertension for 10 years, which was partially controlled by the daily use of 100-mg atenolol.

Systemic work up did not reveal any secondary cause of hypertension; therefore, she was diagnosed with essential hypertension. Brain MRI findings were also normal. At the time of headache, the patient was recommended to use 100% oxygen at a rate of 8 L/min, but it had no effect on the headache and blepharospasm. We also administered two doses of 100-mg sumatriptan at 2-hour intervals, but these were also not effective. Thereafter, Dysport was injected to the periorbital, temporal, and occipital areas in small unit doses, and this resulted in an improvement in blepharospasm and, to some extent, tearing and rhinorrhea after 4 days; however, the headache continued as before [Figure 1b].

Since the headache did not respond to the above-mentioned therapy, we consulted a neurologist and administered a combination therapy with 125-mg sodium valproate two times a day for 1 week, 40-mg propranolol tablets every 12 hours, 25-mg nortriptyline every night, and 4-mg perphenazine every night for 1 month. This combined medical therapy alleviated the headache significantly, and the patient reported a 70% reduction in the recurrence rate and severity of headache over 1 month, as well as a remission of blepharospasm.

After 1 month, the drug dosage was increased to 250-mg sodium valproate every 12 hours, 50-mg nortriptyline every night, and 8-mg perphenazine every night; moreover 50-mg pregabalin tablets every night were added to the treatment regimen. Six months after the initiation of treatment, the patient reported that the severity and recurrence rate of headache was controlled up to 90% and that the blepharospasm did not relapse. Moreover, her anxiety and depression symptoms were considerably improved, and systemic hypertension was well controlled.

DISCUSSION

The periodicity of cluster headache attacks indicates the involvement of a biologic clock within the hypothalamus, with central disinhibition of the nociceptive and autonomic pathways, especially the trigeminal nociceptive pathways.^[11] Cluster headache is a member of the group of disorders called TACs. The trigeminal distribution of pain and ipsilateral autonomic features of these disorders suggest the presence of a trigeminal–autonomic reflex^[12] or trigeminal autonomic activation. Trigeminal and facial nerve communication and their anatomical and functional relationship have been investigated before.^[13,14] Nakazato et al reported the case of a man with typical cluster headache who developed daily morning hemifacial spasm, which occurred with a pattern similar to cluster headache. Therefore, the authors suggested



Figure 1. Patient's face: (a) before Dysport injection showing severe blepharospasm. (b) After Dysport injection showing improvement in blepharospasm.

that hemifacial spasm and cluster headache have the same mechanism.^[11] In the present case, the patient had unilateral blepharospasm with fine orbicularis oculi twitching movement and brow ptosis, but her lower facial muscles were not involved. Another case of a patient with long-lasting blepharospasm, photophobia, lacrimation, and rhinorrhea that did not respond to bilateral Dysport injection was reported by van Vliet et al.^[12]

Our patient had a past medical history of systemic hypertension, and systemic work up revealed the presence of essential hypertension with no secondary cause. A previous study has revealed that both systolic and diastolic blood pressure significantly increased during cluster headache attacks.^[15]

The presence of abnormal face movement, i.e., hemifacial spasm or blepharospasm, was not a typical feature of cluster headache or TACs.^[2,16,17] A literature search revealed only two reports of abnormal face movement in patients with TACs.^[11,12] Our patient had experienced episodes of typical cluster headache and blepharospasm for 10 years, and over time, she developed anxiety, constant headache, and systemic hypertension. This suggests that beside the basic pathophysiology of TACs and trigeminal and facial nerve communication, psychologic factors may be involved in the development of this disease combination. Although an anatomical and functional connection exists between the trigeminal and facial nerves and episodic hemifacial spasms reported in patients with cluster headache have been considered to have the same mechanism as cluster headache,^[11] psychologic factors and the patient's compensatory response to the pain may have an influence of the condition.^[18,19] The patient's symptoms did not respond to conventional treatment for cluster headache but improved up to 90% with a combination of drugs having analgesic, anti-anxiety, and anti-depression effects, as well as prophylactic agents for cluster headache and symmetrical botulinum toxin A injection to the bilateral orbicularis oculi, temporalis, and occipital muscles. Therefore, we conclude that unilateral secondary blepharospasm in this patient with cluster headache had a multifactorial origin. Future research may reveal the exact mechanism or even the prevalence of facial movement disorders in association with cluster headache, and may reveal whether increasing the systolic and diastolic blood pressure during cluster headache attacks may lead to the development of essential systemic hypertension.

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Conflicts of Interest

There are no conflicts of interest.

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