Neurology Asia 2025; 30(1) : 341 – 343

Rare trigeminovascular signs during migraine attack

[1]Aslihan TASKIRAN-SAG, [2]Tugce KABAKCI, [2]Cagla OZDOL

[1]Department of Neurology, Faculty of Medicine, TOBB University of Economics and Technology, Ankara, TURKIYE; [2]Faculty of Medicine, TOBB University of Economics and Technology, Ankara, TURKIYE

Abstract

Severe headache and physical findings in the face and neck area may imply serious intracranial pathology and warrant further investigations including laboratory tests and imaging. We present two migraine cases with unusual trigeminovascular cutaneous findings (i.e., periorbital bruising and facial swelling). Recognition of these rare manifestations of migraine may help the clinician in differential diagnosis and prevent unnecessary investigations. A pathophysiological discussion of these signs is provided.

Keywords: Migraine, facial swelling, ecchymosis, trigeminovascular system, skin findings

INTRODUCTION

Migraine is a complex neuro-glio-vascular headache disorder with a genetic predisposition. Although more typical for trigeminal autonomic cephalalgias, additional symptoms such as conjunctival injection, lacrimation, flushing, pupil changes, and sweating may also occur during migraine headaches. However, cutaneous findings are rare among migraine patients. Here, we present two cases with unusual trigeminovascular cutaneous manifestations that occurred during migraine attacks.

CASE REPORTS

Patient 1

A 36-year-old female presented to the neurology department with a four-year history of recurrent headaches, recently increasing in frequency to seven days per month. The pain was predominantly right-sided. Triggering factors included exposure to spotlights and sunlight, hunger, insomnia, fatigue, and menstruation. During attacks, she occasionally experienced transient numbness on the right side of her face and mild dysphasia. Her most recent headache episode began the day before her initial presentation. She described the headache as severe (7 out of 10 on the visual analog scale), bilateral, and more intense periorbitally. There was no preceding aura. The headache was more severe on the right side and was accompanied by photophobia, phonophobia, peripheral sensitivity, and nausea. She also noticed swelling on her forehead and upper lip lasting 6–7 hours, which she had experienced a few times during previous headache attacks (Figure 1a). She experienced partial relief with flurbiprofen.

The patient was not using combined oral contraceptives and had no other illnesses. There was a family history of migraine in her mother and sister.

On physical examination, her vital signs were within normal limits, and the swellings had resolved completely. The results of a neurological examination were unremarkable, and blood tests did not reveal any abnormalities. An MRI scan showed two small nonspecific hyperintense lesions in the frontal white matter.

The patient was diagnosed with migraine, and flunarizine was prescribed prophylactically. She was referred to the ophthalmology and ear, nose, and throat departments to rule out other conditions that might have caused local swelling on the face, but no other illnesses were found.

Patient 2

A 37-year-old female presented to the emergency department with a unilateral, moderate to severe left-sided throbbing headache, accompanied by ipsilateral periorbital ecchymosis and ipsilateral facial numbness. She reported that the ecchymosis and facial numbness appeared before the headache, with the numbness lasting 15 minutes. She had been working on a computer for an

Address correspondence to: Aslihan TASKIRAN-SAG, Department of Neurology, TOBB ETU Faculty of Medicine Hospital, Yasam cd, no: 5, Bestepe, 06560, Ankara, TURKIYE. Email: aslihantaskiran@gmail.com

Date of Submission: 2 July 2024; Date of Acceptance: 12 November 2024

https://doi.org/10.54029/2025sni

extended period of time before the headache started. Her blood pressure, heart rate, and body temperature were normal, and physical examination was unremarkable except for a small ecchymosis on her left lower eyelid (Figure 1b). She had no history of systemic illnesses such as diabetes or hypertension. After the exclusion of a neurovascular emergency, she was referred to the neurology department.

During the neurological evaluation, the patient reported a history of unilateral headaches during her menstrual cycles; however, this was the first time she experienced a headache with cutaneous symptoms. Her mother had also experienced unilateral recurrent headaches (without any skin manifestations) at a younger age. The patient reported no visual symptoms, photophobia, phonophobia, irritability, or malaise. Both the neurological examination and ophthalmological assessment yielded normal results. Blood tests revealed mild deficiencies of vitamin D and ferritin. Her coagulation profile was normal.

A magnetic resonance scan was performed, and the results were normal. The clinical diagnosis was migraine with aura.

DISCUSSION

Trigeminovascular system activation is proposed as a final common pathway in the pathogenesis of migraine attacks.[1] Upon activation of the trigeminal afferents surrounding pial and dural vessels, signals pass through the trigeminal ganglion to the trigeminal nucleus caudalis. The projections of second-order trigeminal neurons are extensive and mediate both pain generation and the autonomic and affective symptoms accompanying a migraine attack.[1] The trigeminal nerve endings contain and release several compounds. The action of these mediators can cause vasodilation, plasma protein leakage, tissue edema, and mast cell degranulation, inducing neurogenic inflammation in target tissues, including the meninges and brain parenchyma.

In our Patient 1, the facial swelling occurred during the headache phase and lasted 6–7 hours. This may be related to the release of neuropeptides during the late phase of trigeminovascular activation which resulted in inflammation. The trigeminal nerve also supplies cutaneous innervation to the face, and similar mechanisms to those in the meninges might operate in the extracerebral area supplied by the trigeminal nerve.

Cerebral and dural blood flow changes during migraine attacks are well known. The middle meningeal artery and dural vessels dilate in the early phase of a migraine attack. However, it is unclear whether the cutaneous vasculature of the facial area undergoes vasodilatation. Our Patient 2 presented with a throbbing headache and a small periorbital bruise on the left side, along with a sensory aura. Since the ecchymosis and the facial numbness appeared spontaneously before the headache, we speculate that these symptoms were manifestations of the aura phase or early activation of the trigeminovascular system.

In the literature, there are a few similar cases of migraine as well as a case of atypical paroxysmal

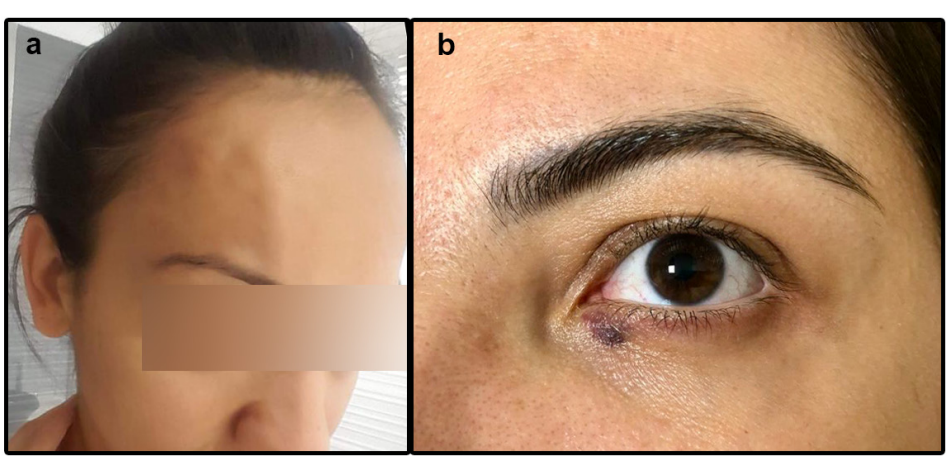

Figure 1. The picture in panel a was acquired by Patient 1 at home during a previous migraine attack. The image demonstrates areas of patchy swelling in the right V1 distribution at the patient's forehead. The picture in panel b was taken by the neurologist during the examination of Patient 2 for headache. It demonstrates a purpuric macula on the lower eyelid ipsilateral to the headache.

headache associated with facial ecchymosis.[2-5] Some of these cases are referred to as red forehead dot syndrome. In a study by Durmaz *et al*., punch biopsy from the red skin lesions that appeared during a migraine attack revealed perivascular dermatitis, abundant plasma cells, and extravasated erythrocytes in the dermis.[6] This suggests a perivascular inflammatory response that may be due to the action of trigeminovascular neuropeptides. However, in our second case, the lesion was a single ecchymosis rather than an erythematous lesion. Therefore, it might have resulted from aberrant vasodilation occurring with the trigeminal autonomic reflex activation.[1] Occasional cases of epistaxis accompanying migraine may also support the extracranial vasomotor changes caused by trigeminovascular activation.[7,8]

In conclusion, recognition of these rare manifestations of migraine may help clinicians in differential diagnosis and prevent unnecessary investigations.

DISCLOSURE

Ethics: Informed consents have been obtained from the patients.

Financial support: None

Conflict of interest: None.

REFERENCES

1. Ashina M, Hansen JM, Do TP, Melo-Carrillo A, Burstein R, Moskowitz MA. Migraine and the trigeminovascular system-40 years and counting. *Lancet Neurol* 2019;18:795-804. doi: 10.1016/S1474-4422(19)30185-1.
2. DeBroff BM, Spierings EL. Migraine associated with periorbital ecchymosis. *Headache* 1990;30:260-3. doi: 10.1111/j.1526-4610.1990.hed3005260.x.
3. Sethi PK, Sethi NK, Torgovnick J. Teaching NeuroImages: Red forehead dot syndrome and migraine revisited. *Neurology* 2015;85:e28. doi: 10.1212/WNL.0000000000001762.
4. Monaghan BM, Smyth S. Teaching NeuroImages: Red forehead dot syndrome and migraine revisited. *Neurology* 2016;86:697-8. doi: 10.1212/01.wnl.0000481056.72967.7a.
5. Comabella M, Titus F, Huguet P. Recurrent paroxysmal headache associated with facial ecchymosis. *Cephalalgia* 1996;16:341-3. doi: 10.1046/j.1468-2982.1996.1605341.x.
6. Durmaz EO, Demirkesen C. Red forehead dot(s) and migraine. *Indian J Dermatol Venereol Leprol* 2020;86:704-5. doi: 10.4103/ijdvl.IJDVL_177_20.
7. Peretz AM, Woldeamanuel YW, Rapoport AM, Cowan RP. Spontaneous extracranial hemorrhagic phenomena in primary headache disorders: A systematic review of published cases. *Cephalalgia* 2016;36:1257-67. doi: 10.1177/0333102415618951.
8. Bhatia S, Bahl VJ, Sharma M, *et al*. Recurrent Epistaxis: An unusual manifestation of clinical spectrum of migraine-Case series with scoping review of literature. *Indian J Otolaryngol Head Neck Surg* 2023;75:984-93. doi: 10.1007/s12070-023-03542-6.