Dear Editor,

Epicrania fugax (EF) was first described by Pareja et al in 2008 [1]. It has been recently included in the Appendix of the 3rd edition of The International Classification of Headache Disorders (ICHD-III, beta version) [2]. It is characterized by ultrabrief paroxysms of unilateral pain spreading from the posterior scalp to the ipsilateral eye or nose, along a linear or zigzag trajectory, and lasting less than 10 s. Pain intensity can be moderate or severe, and pain quality has been described as electric or stabbing. Autonomic symptoms can be present. Pain frequency is extremely variable and most attacks are spontaneous, but some of them can be provoked by tactile stimulation. Between paroxysms, mild pain or hyperesthesia could remain in the stemming area but complete disappearance of pain is also possible [1].

As the first description of EF, there have been several publications supporting this new entity. A total of 66 cases have been reported up to now, 48 of them sharing the original forward radiation. Conversely, a backward radiation from frontal or periorbital areas toward the occipital region has been described in 18 cases [3,4]. Pain paroxysms are strictly unilateral, although some patients have shifting sides [3]. Associations with other headaches, mainly nummular headache (NH), cluster headache or migraine, have been reported [5]. The pathogenesis of this new entity is still a subject for discussion.

We wish to describe the first case with clinical features similar to EF whose pain is strictly located in the mid sagittal plane. A 47-year-old man came to our headache clinic complaining of a 3-month history of paroxysmal head pain. Pain paroxysms always started at the same point in the occipital sagittal scalp, and rapidly moved forward in a linear trajectory along the sagittal suture up to the space between the eyebrows. Duration of the complete sequence was very brief, hardly 3 s, and the average frequency was six attacks per day. Pain quality was described as stabbing, and its intensity was rated as 7 out of 10 in a visual analogue scale (0: no pain, 10: the worst imaginable pain). There were neither autonomic symptoms nor interictal pain. The patient did not describe any triggers. When questioned about other headaches, the patient revealed a history of chronic migraine for several years, without aura and currently well controlled without preventive treatment. Pain was fronto-parietal, side-shifting, and considered by the patient as quite different from this new one. General and neurological examinations were normal. Palpation of supraorbital, infraorbital, supratrochlear, minor occipital, and greater occipital nerves caused no pain.

We did not obtain pathological findings in routine blood work-up, immunological screening, or magnetic resonance imaging. Once the patient was informed about the benign nature of his syndrome, he considered the pain not annoying enough to require preventive therapy.

We hereby present a case report of a patient with brief and dynamic characteristics considered distinctive of EF, with a not previously described pain topography. Pain always stemmed from the median occipital region of the scalp, and rapidly moved forward to the space between the eyebrows, with a midline trajectory along the sagittal suture. The pathogenesis of EF remains uncertain. The localized onset of pain, its stabbing character and the occasional symptoms and signs of sensory dysfunction on the stemming area suggest an epicranial source [6]. Pain paroxysms have been thought to initiate in terminal twigs of the pericranial nerves. It has been proposed that the spreading from onset to the end is caused by aberrant ephaptic transmission through different nerve fibers, by transdiploic transmission [1], or even by central mechanisms [7]. The present description encourages us to carefully analyze the differences between EF and epicranial neuralgias, as they are also characterized by a dynamic paroxysmal pain along the nerve territory. Occipital neuralgia has been described as pain originating from the posterior neck that often radiates to the eyes. However, the anatomy of the occipital nerves does not explain radiations beyond the vertex. Given the anatomical convergence of cervical and trigeminal afferents at the trigeminal nucleus caudalis, a central mechanism could explain it [8–10]. An involvement of the lesser occipital nerve, a direct branch of dorsal root C3, which inervates the skin of medial occipital bone [8], may be found in this patient. Nevertheless, pain in midline, anterior to occipital, cannot be attributed to any occipital nerve. If we consider forehead pain, supraorbital neuralgia or neuralgia of V1 can be other possibilities. However, trigeminal neuralgia is usually more severe and supraorbital neuralgia is characterized by continuous pain with no exacerbations or neuropathic symptoms. Neither of them explain the midline distribution unless there was a bilateral lesion. Moreover, there was no tenderness of epicranial nerves, which is typical in neuralgias. As for NH, it is the most typical epicranial disorder. NH is defined as a continuous or intermittent pain in a small circumscribed rounded or elliptical area of the scalp, in the absence of any underlying structural lesion [2]. Sagittal variants of NH have been described and exacerbations are also possible [9]. However, despite sharing a focal onset of the pain, in EF it has a dynamic character, while in NH the pain begins and ends in the same
area. Because of the autonomic signs, EF could resemble SUNCT (Shortlasting Unilateral Neuralgiform headache attacks with Conjunctival injection and Tearing) or SUNA (Shortlasting Unilateral Neuralgiform headache attacks with cranial Autonomic Features). Both consist in paroxysms of neuralgiform periorbital pain with prominent lacrimation or conjuntival injection. Several cases of EF with autonomic signs have been reported, but they are not present in our patient.

In conclusion, our patient fulfills essential features of EF, but it is the first case with sagittal localization so far. This topography does not fit well with other known headaches or neuralgias. The pain may be attributed to terminal sensitive branches instead of a nerve trunk. The dynamic component of the pain from the onset to the end may be explained by aberrant ephatic transmission through different nerve fibers, or either by transdiploic transmission. Central mechanisms can also explain the spreading of pain, through anatomical convergence of cervical and trigeminal afferents at the trigeminal nucleus caudalis.

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References


Low-Dose Naltrexone to Prevent Intolerable Morphine Adverse Events: A Forgotten Remedy for a Neglected, Global Clinical Need

Dear Editor,

Opioids are the mainstay of treatment for moderate-severe pain but their use is dramatically limited by intolerable adverse events (AEs), such as constipation, nausea, vomiting, somnolence, dizziness, pruritus, and urinary retention [1,2]. Although opioid AEs constitute a long-standing clinical problem, effective treatment strategies are still not available. Thus, to date opioid AEs continue to represent an insurmountable barrier to therapy for millions of sufferers, who are denied the chance to receive an adequate pain relief.

Even though symptomatic drugs are used to treat opioid AEs, evidence supporting their efficacy is very low and, moreover, the management of several AEs with multiple adjuvant agents increases the risk of harmful drug interactions and diminishes compliance [2]. Although the opioid rotation method proved some efficacy in reducing AEs, this approach requires highly specialized