

recognized statistical method to report odds and risk relationships in large nationally representative data sets. Finally, as an additional precaution, we made clear in our *Discussion* section that cross-sectional survey data cannot be used to make causal statements but only highlight potential areas for future research. We thank Dr. Woldeamanuel for reinforcing these important points and the need for more research.

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Letter to the Editor

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Epicrania Fugax with a Novel Sign: Pain Paroxysms with Parallel Forward or Backward Trajectories

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Dear Editor,

Epicrania fugax (EF) manifests as ultrabrief electric or stabbing pain paroxysms on the cranium surface of moderate or severe intensity. Sometimes, patients with EF also experience ocular or nasal autonomic accompaniments and interictal mild pain or hyperesthesia [1]. The frequency of these paroxysms varies greatly, from a few attacks per year to numerous attacks per day. Mostly, the paroxysms occur spontaneously, but they can also be triggered by touch or palpation. First reported by Pareja et al. in 2008 [2], EF has since been listed in the Appendix of the 3rd edition of the International Classification of Headache Disorders (ICHD-3) (Table 1) [3].

EF patients display various pain features that stem from posterior cranial areas moving to the ipsilateral anterior scalp, or from the frontal areas, eye, or nose toward the parietal or occipital region along lineal or

zigzag trajectories [4, 5]. We performed a retrospective study on the clinical course of 960 patients attending a headache specialist clinic from January 2016 to December 2017 in our hospital. Among these patients, the three cases meeting diagnostic criteria for EF with a specific pain radiating pattern were reviewed.

The first patient was a 62-year-old woman who complained of headache paroxysms for three months. The pain was described as electric, with an intensity of 5 out of 10 on a visual analog scale (VAS; 0 = no pain, 10 = the worst imaginable pain). The paroxysms began in the right eye and spread posteriorly and caudally along two parallel backward zigzag trajectories to reach the ipsilateral suboccipital region and lasted for three seconds every time (Figure 1A). These attacks happened around twice per month and showed no apparent triggers. Physical or neurological examinations, routine blood tests, and a magnetic resonance imaging (MRI) scan of the brain detected

Table 1. Diagnostic criteria for epicrania fugax (ICHD-3)

A4.11 Epicrania fugax

Description:

Brief paroxysmal head pain, with stabbing quality, describing a linear or zig-zag trajectory across the surface of one hemicranium

Diagnostic criteria:

A. Recurrent stabbing head pain attacks lasting 1–10 seconds, fulfilling criterion B

B. The pain is felt to move across the surface of one hemicranium in a linear or zig-zag trajectory, commencing and terminating in the territories of different nerves

C. Not better accounted for by another ICHD-3 diagnosis

ICHD-3 = 3rd edition of the International Classification of Headache Disorders.

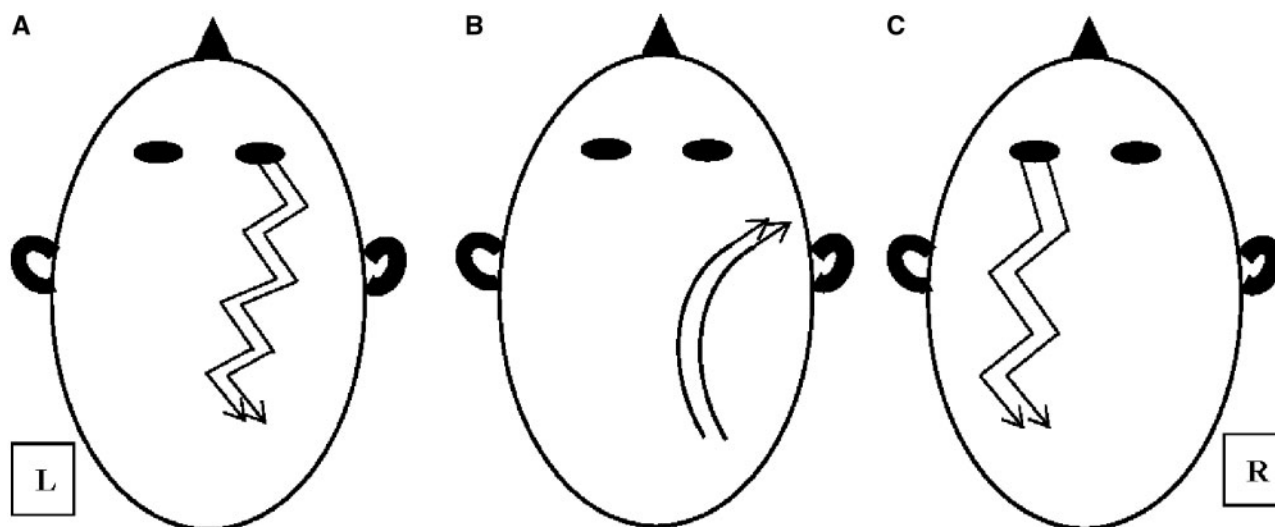


Figure 1. Moving pattern of pain in the three patients. A) Right eye to ipsilateral suboccipital region in two parallel backward zigzag trajectories. B) Right occipital area to right temporal scalp in two parallel forward linear trajectories. C) Left eye to ipsilateral suboccipital area in two parallel backward zigzag trajectories.

no abnormalities, but paresthesia along the spreading route of the pain was obvious. The patient rejected any preventive therapy and maintained similar pain quality, intensity, and frequency during the one-year follow-up.

The second patient was a 42-year-old man who was referred to our clinic due to headache paroxysms for three years. The pain had a stabbing quality, an intensity of 6 out of 10 on the VAS, and started in the right occipital area, moving anteriorly in two parallel linear trajectories to the right temporal scalp (Figure 1B). Attacks occurred two to three times per week and lasted about 10 seconds. Touching the scalp triggered the pain paroxysms, but they also occurred spontaneously. Moreover, a mild interictal pain at the starting site was mentioned. Physical and neurological examinations, routine blood tests, and brain MRI were normal. After treatment with carbamazepine (100 mg, bid) for one month, the patient stopped treatment and reported no attacks during the subsequent 11-month follow-up.

The third patient was a 56-year-old woman who consulted us for intermittent headache paroxysms of five months' duration. The pain was described as electric, starting from the left eye and moving caudally in two parallel zigzag trajectories to the ipsilateral suboccipital

area (Figure 1C), lasting five seconds each time. The pain intensity was rated as 5 out of 10 on the VAS, and the episodes occurred three times per week. The patient showed no symptoms between paroxysms, although a history of migraine without aura was declared. Physical and neurological examinations, routine blood tests, and brain MRI were all normal, as in patients 1 and 2. We treated the patient with carbamazepine (100 mg, bid) for one month, the attacks stopped, and treatment was discontinued. During the one-year follow-up after discharge, no relapse was reported.

Here we introduce three cases of EF with pain spreading along two parallel trajectories, a pattern never before reported. EF was originally regarded as a pain paroxysm spreading from posterior cranial areas to the ipsilateral anterior scalp in a single linear or zigzag trajectory [2], after which EF with backward radiation was proposed in 2010, 2011, and 2013 [4, 6, 7]. Recently, four EF patients with pain spreading both forward and backward in the same person were reported [5]. In addition, pain traversing the midline to the contralateral side [2, 7], radiating coronally [8], or moving in multiple directions [9] has been described. But our cases are the first reported in Asian patients. The diverse pattern of pain radiation

indicates the potentially complex mechanisms underlying the pathogenesis of EF.

Mechanisms underlying the etiology of EF include aberrant ephaptic transmission connecting different nerve fibers, transdiploic conductance of signals through pain fibers, and functional convergence of different afferents within the central nervous system [10]. Furthermore, central mechanisms could explain the advent of autonomic symptoms. The patterns of paroxysms in our three cases are consistent with the potential contribution of the supraorbital nerve in ophthalmic division of the trigeminal nerve (V-1) and the greater occipital nerve in the second cervical root (C2).

In conclusion, we reported three cases fulfilling the diagnostic criteria of EF but with pain spreading in a distinctive manner, which might represent a new variant of EF. Our description might help enrich the diverse clinical phenotypes of EF and reinforce the differential diagnosis between EF and other primary headaches.

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Letter to the Editor

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Ultrasound Imaging for Recalcitrant Lateral Elbow Pain: Radio-Humeral Synovial Plica Is Also at Play

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