LETTER TO THE EDITOR **Open Access**

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Postictal Prosopometamorphopsia after Focal Status **Epilepticus due to Cavernous Hemangioma** in the Right Occipital Lobe

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Dear Editor,

Prosopometamorphopsia (PM) is the visual distortion of face perception, which is generally accompanied by other types of visual illusions. It is mainly associated with the fusiform face area and the adjacent occipital face area. There are rare reports of PM in patients with stroke, epilepsy, tumor, and migraine, and also in patients with eye disease. 1 Cases involving epilepsy have only been reported as ictal manifestations.² Herein we describe a man with variable manifestations of postictal PM without other visual illusions after focal status epilepticus in occipital lobe epilepsy.

A 45-year-old man presented with a 1-week history of visual disturbances. He experienced intermittent flashes of light with blurring of vision in the left visual field. He had a history of generalized tonic-clonic convulsions in 2004. At that time he was diagnosed with cavernous hemangioma in the right occipital area. He responded favorably to valproate, experiencing no symptoms while receiving this treatment for 10 years. However, he had stopped taking valproate on his own 2 years previously. Neurologic examination was normal except for left homonymous hemianopsia. Contrast-enhanced MRI of the brain including T1-weighted, T2 weighted, fluid-attenuated inversion recovery, and gradient-echo sequences showed unchanged cavernous hemangioma from that in the previous study, but was otherwise normal. Electroencephalogram (EEG) revealed repeated periodic rhythmic fast activities confined to the right occipital area that were suggestive of focal status epilepticus (Supplementary Fig. 1 in the online-only Data Supplement).

Intravenous valproate was administered as a loading dose of 20 mg/kg, followed by 450 mg of oral valproate twice daily. His visual symptoms disappeared over the next 2 days. However, on day 3 he started visualizing distortion of people's faces, describing that they looked like a two-dimensional plane. The distortion was mainly in the right half of the face, and the right eye and the chin appeared wavy with a blurred image of the nose. His visual impairment was specific to face perception only. The patient's drawing of his doctor's face depicted PM over the entire face, which it being more severe in the right half (Fig. 1A). Follow-up EEG showed well-developed and symmetric posterior alpha activity without focal slowing. Accordingly, postictal PM was strongly suspected. On day 4 his symptom partially improved, with another drawing of his doctor's face now showing PM restricted to the right half of the face (Fig. 1B). The patient was discharged on day 5, at which time his symptom had not completely resolved. At the 1 week follow-up the patient reported that he no longer saw distorted faces of people, and a third drawing depicted his doctor's face normally (Fig. 1C).

Until now, PM has been reported as an ictal manifestation. Our patient developed postictal visual illusion restricted to the face only, which has not been reported in cases of epilepsy. Although the exact mechanism of PM remains unclear, the fusiform face area and occipital

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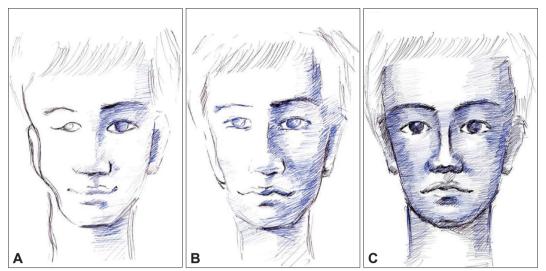


Fig. 1. Patient's drawing of his doctor's face. A: A drawing made on day 3 depicts bilateral prosopometamorphopsia (PM) that is more severe in the right half of the face. B: A drawing made on day 4 depicts unilateral PM restricted to the right half of the face. C: A drawing made on day 11 depicts a normal face.

face area are considered to play a major role.³ It is hypothe-sized that the occipital face area of each hemisphere recognizes the contralateral side of the face, and that facial information from both hemispheres is transmitted to the fusiform face area in the right hemisphere. It is interesting that postictal cerebral dysfunction of right occipital area in our case initially led to bilateral PM, which was subsequently changed into a unilateral pattern. This is strongly suggestive that PM can present with variable phenotypes based on the pattern of involvement of the right occipital lesion, and also supports the above hypothesis that facial information from both hemisphere is transmitted to the right occipital area.

Supplementary Materials

The online-only Data Supplement is available with this article at http://dx.doi.org/10.3988/jcn.2016.12.3.371.

Conflicts of Interest _

The authors have no financial conflicts of interest.

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