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Case Report

A case of confusional migraine with transient increased cerebral blood flow

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Abstract

Background: Confusional migraine is a rare type of migraine presenting as an acute confusional state. However, the mechanism of this confusional state remains unclear.

Subject and methods: We examined an 11-year-old girl with confusional migraine, using electroencephalography, brain magnetic resonance imaging, cerebrovascular magnetic resonance angiography, and single-photon emission computed tomography to investigate cerebral blood flow changes.

Results: Our findings revealed vessel narrowing in the left middle and posterior cerebral artery territory, indicating vasospasm and suggesting that the confusion was caused by hypoperfusion. However, abnormal increased cerebral blood flow in the left middle and posterior cerebral artery territory was observed during the non-confusional state.

Conclusion: The recorded cerebral blood flow changes are similar to those associated with migraine attacks, gradually changing from abnormally low to abnormally high during the confusional and post-confusional state.

Keywords: Confusional migraine; Cerebral blood flow; Electroencephalography; Magnetic resonance angiography; Single-photon emission computed tomography

1. Introduction

Confusional migraine was first reported by Gascon et al. in 1970 [1] as a rare type of migraine that presents as an acute confusional state. The mechanism of the confusional state remains unclear, although previous reports suggest that it is caused by cerebral ischemia with vasospasm of the cerebral artery [2,3]. Here, we report a case of confusional migraine, in which we studied cerebral blood flow changes using electroencephalography (EEG), brain magnetic resonance imaging (MRI), cerebrovascular magnetic resonance angiography (MRA), and single-photon emission computed tomography (SPECT).

2. Case report

The patient was a right-handed girl aged 11 years, 10 months. From the morning of admission, the patient experienced numbness spreading from the right upper extremity to the right corner of the mouth, right visual

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field disturbance, vomiting, and headache. She gradually became unable to communicate due to disturbed consciousness and restlessness, and was admitted to our hospital. Her family history included her mother, who had a history of pulsating headaches. There was no family history of febrile seizures or epilepsy.

The patient had experienced repeated numbness on the right side of her body, two or three times a year from the age of 7 years, 5 months. She was admitted to our hospital at 7 years, 8 months with numbness of the right body and a confusional state. Consciousness returned to normal on the second day after onset. EEG findings on the third day after onset were normal. However, 99mTc-ethylcysteinate dimer (ECD)-SPECT findings on the fourth day after onset revealed mild hyperperfusion in the left cerebral hemisphere, with epilepsy suspected. She was followed up without administration of antiepileptic drugs. She had no history of major or minor head injuries. Her birth and developmental history were normal.

On admission, the patient had a body temperature of 36.8 °C, heart rate of 90 beats/min, blood pressure of 108/60 mmHg, and respiratory rate of 24 breaths/min. She exhibited delirium with a Glasgow Coma Scale score of E2 V4 M5. She was in a confused state, and was unresponsive to commands, restless, and disorientated. Neurological examinations were normal except for this reduced level of consciousness.

Right hemi-numbness, visual disturbance, vomiting, and headache appeared 1 or 2 h after onset, and consciousness gradually deteriorated to delirium and restlessness 2 h after onset. The patient was initially unable to move the extremities because she was experiencing confusion, and grip strength in the right hand was weakened because of a sensation of numbness. Headache, nausea, consciousness, and right grip strength had returned almost to normal after approximately 8 h, and had resolved completely by the following day. However, the patient remembered few details of the confusional episode.

Laboratory blood test results were normal. Cerebrospinal fluid analysis showed mildly elevated protein (450 mg/L) without pleocytosis or reduced glucose. Cerebrospinal fluid lactate and pyruvate levels and the lactate/pyruvate ratio were within the normal range.

Brain MRI on the day of onset showed no abnormalities. Cerebrovascular MRA was also performed during the confusional episode at 6 h after onset (Fig. 1a), with narrowing of the left middle cerebral arteries detected. However, MRA images at 1 month after onset (obtained without any confusional episode) showed no vascular stenosis or occlusions (Fig. 1b).

EEG during the confusional episode at 3 h after onset showed diffuse left-side dominant high-voltage slow waves, which were particularly marked in the left frontal area during awake recordings (Fig. 2a). EEG on the third day after onset did not show slow or seizure waves (Fig. 2b).

99mTc-ECD-SPECT on the second day after onset showed hyperperfusion in the left temporal to occipital cerebral regions. (Fig. 3a). By the tenth day (Fig. 3b), there was no difference in cerebral blood flow between the left and right hemispheres.

One and a half months after discharge from hospital, the patient was hospitalized again with a similar confusional episode. She had a total of five confusional episodes from the age of 7 years and 8 months. Consequently, sodium valproate (VPA) was initiated to prevent further episodes. The confusional migraine attacks were completely controlled by 500 mg/day of VPA.

3. Discussion

The diagnostic criteria for confusional migraine were proposed by Pietrini et al. in 1987 [4], and further details were added by Pacheva and Ivanov in 2013 [5]. In the present case, all diagnostic criteria [4,5] were satisfied, except for the absence of significant neuroimaging abnormalities: brain SPECT images revealed abnormal findings in our patient. Thus, confusional migraine was diagnosed.

According to Olesen et al., during migraine attacks, regional cerebral blood flow in the posterior cerebral hemisphere generally decreases during the aura phase and gradually changes from abnormally low to abnormally high during the headache phase [6]. Nezu et al., reported that brain ^{123}I-IMP-SPECT performed within 48 h of confusional migraine attacks revealed hypoperfusion in the left posterior cerebral artery territory [2]. Fujita et al., also reported that intracranial MRA during confusional migraine attacks indicated transient narrowing of the left middle and posterior cerebral arteries [3]. These findings provide evidence of vessel narrowing in the middle and posterior cerebral artery territory, indicating vasospasm, suggesting that the confusion is due to hypoperfusion. Miyake reported a girl aged 7 years and 2 months with confusional migraine, exhibiting a different pattern of results [7]. Although EEG during the confusional state showed slow wave activity and an absence of ipsilateral sleep spindle activity in the left hemisphere, ECD-SPECT revealed hyperperfusion in the left hemisphere with no confusion 24 h after onset [7].

In the present case, at both 7 years, 8 months and this time, brain 99mTc-ECD-SPECT images were acquired during the non-confusional state on the second and fourth days after the confusional migraine attack, respectively. These images also showed hyperperfusion in the left cerebral hemisphere. These findings are very similar to those of Miyake's study [7]. The hyperperfusion findings of our regional cerebral blood flow

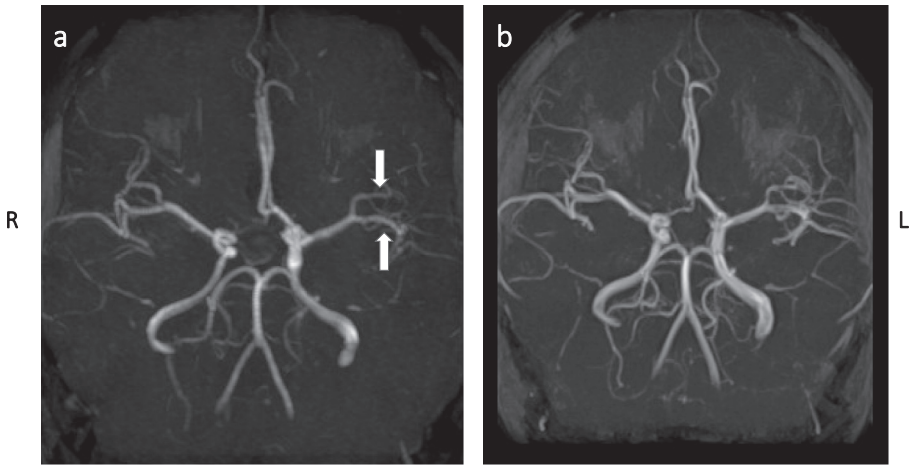

Fig. 1. Cerebrovascular magnetic resonance angiography (MRA) findings. (a) MRA image during a confusional episode 6 h after onset. Arrows show narrowing of left middle cerebral arteries. (b) MRA image with no confusional episode at 1 month after onset. There was no vascular stenosis or occlusion findings (right).

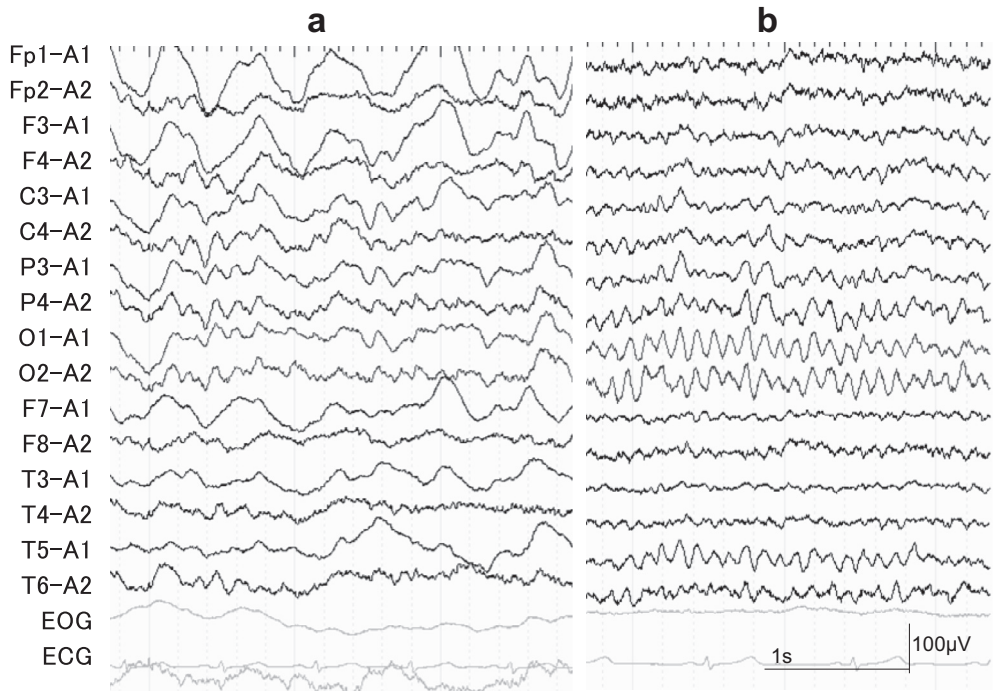

Fig. 2. Electroencephalography (EEG) findings. (a) Awake EEG during a confusional episode 3 h after onset, showing diffuse left-side dominant high-voltage slow waves that were particularly marked in the left frontal area. (b) Awake EEG on the third day after onset showing no slow waves or seizure waves.

imaging, performed the day after the confusional attacks using SPECT, indicate increased cerebral blood flow after the event. This suggests that regional cerebral blood flow in confusional migraine is similar to that in migraine attacks [6], gradually changing from abnormally low to abnormally high during the confusional and post-confusional state.

Migraine attacks with typical aura commonly last between 5 and 60 min, during which EEG activity remains normal [8]. In most reported cases of migraine with typical aura, hypoperfusion has been observed to occur during the attack, appearing very soon after aura. However, this hypoperfusion is typically under the threshold of ischemia, and is not sufficiently severe to cause clinical symptoms. Parain et al. reported that EEG during such attacks revealed unilateral slow-wave activity near the temporo-occipital area, with hypoperfusion observed in the same area [9]. Thus, confusional

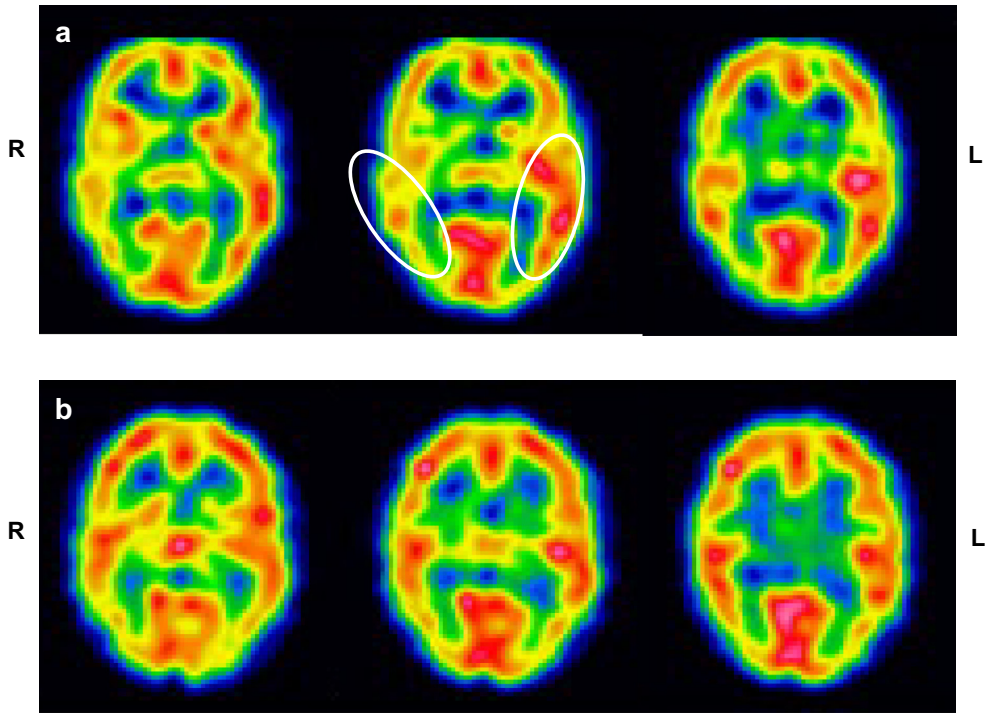

Fig. 3. Brain 99mTc-ethylcysteinate dimer (ECD)-single-photon emission computed tomography (SPECT) images, (a) ECD-SPECT on the second day after onset showing hyperperfusion in the left temporal to occipital cerebral regions. (b) ECD-SPECT on the tenth day after onset showing no clear difference in cerebral blood flow between left and right hemispheres.

migraine with frontal dominant generalized slow waves is thought to involve more severe hypoperfusion than migraine with aura. Based on these findings, we speculated that frontal dominant slow-wave activity caused the confusion state in the current case, whereas occipital-dominant slow-wave activity caused visual aura.

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Potential conflict of interest report

The authors indicated no potential conflict of interest.

Author contributions

E.M, Y.K, K.K, W.I, and A.F. provided medical care and contributed to preparation of the manuscript; E.M. and T.F. drafted the manuscript; T.F. designed the manuscript outline and provided conceptual advice; Y.F. and I.M. supervised the entire study. All authors read and approved the final manuscript.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.braindev.2019.02.002.

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