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A case report : Isolated bilateral midbrain infarction in a healthy female adolescent

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Introduction

Midbrain is a part of brainstem responsible for movement control, arousal, visual processing. Also several nerve fiber tracts and nuclei of the cranial nerves are located in the midbrain. In case of midbrain infarction, due to the anatomical nature of the small vessels, clinicians often infer the causative vessels through ischemic territory and clinical features. The Artery of Percheron (AOP) is known to be a variant of cerebral circulation, a solitary trunk originating from the unilateral P1 segment of posterior cerebral artery (PCA) that supplies to bilateral thalami and midbrain. Ischemic insults at this vessel affect bilateral thalami in most patients and often accompany midbrain infarction. We present an extremely rare case of bilateral midbrain infarction in a healthy 17-year-old female without both thalami involvement and report the mechanism of successful treatment based on neuroanatomical physiology.

Case report

A 17-year-old female visited the emergency room complaining acute onset drowsiness, gait disturbance, mutism and ptosis. On physical examination, postural instability, rigidity in all limbs and extraocular movement limitations were observed. Stroke-related risk factors and family history were denied by her parents. However, magnetic resonance image (MRI) of brain revealed acute infarction of both midbrain. The right P1 segment was invisible and the right P2 segment was well seen branching from the right PCA. (Figure 1.) She was referred to our rehabilitation clinic on the 3rd day of onset. She showed a slow but reproducible response to stimuli and could ambulate requiring moderate assist. However, on the 7th day of onset, bradykinesia, freezing, rigidity and upward gaze limitation especially got worse. Also, she could obey verbal command by non-verbal expression, while initiation of speech and vocalization was impossible. Considering the location of infarction, presenting symptoms were judged to be the result of impairment of the dopaminergic pathway such as bilateral substantia nigra and red nuclei in addition to the lesion of nuclei of 3rd cranial nerve. On the 14th day of onset, levodopa was tried and she recovered alertness on the day 2 of drug administration. Dramatic effect on Parkinsonism and significant improvement on the trail making test were notable. (Figure 2.) The patient has been maintaining the therapeutic effect improving activities of daily living.

Discussion

In this case, considering bilateral midbrain infarction sparing both thalami, occlusion is expected to occur only in the branch of AOP that supplies bilateral midbrain. The cause of bilateral isolated midbrain infarction was difficult to figure out but dramatic effects of levodopa on the characteristic symptoms of Parkinsonism were obvious. However, it

was unclear whether dopaminergic agents on mutism due to ischemic injury of periaqueductal gray matter of midbrain are effective. Follow up of her symptoms should be required.

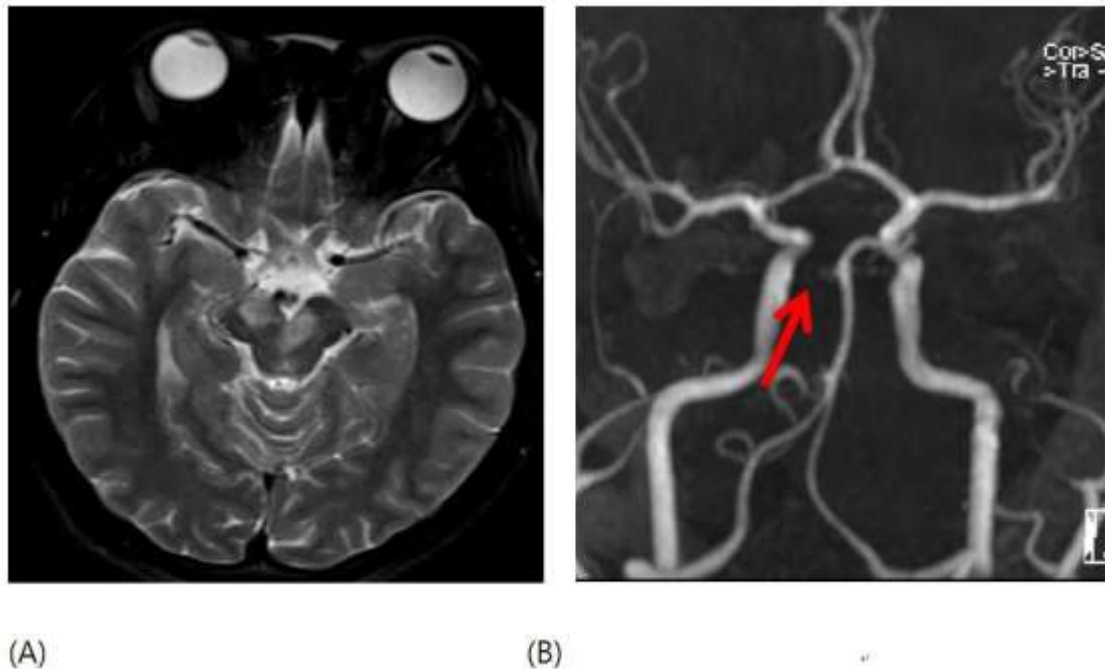


Fig 1. Acute infarction of both midbrain; (A) T2WI; (B) Angiography

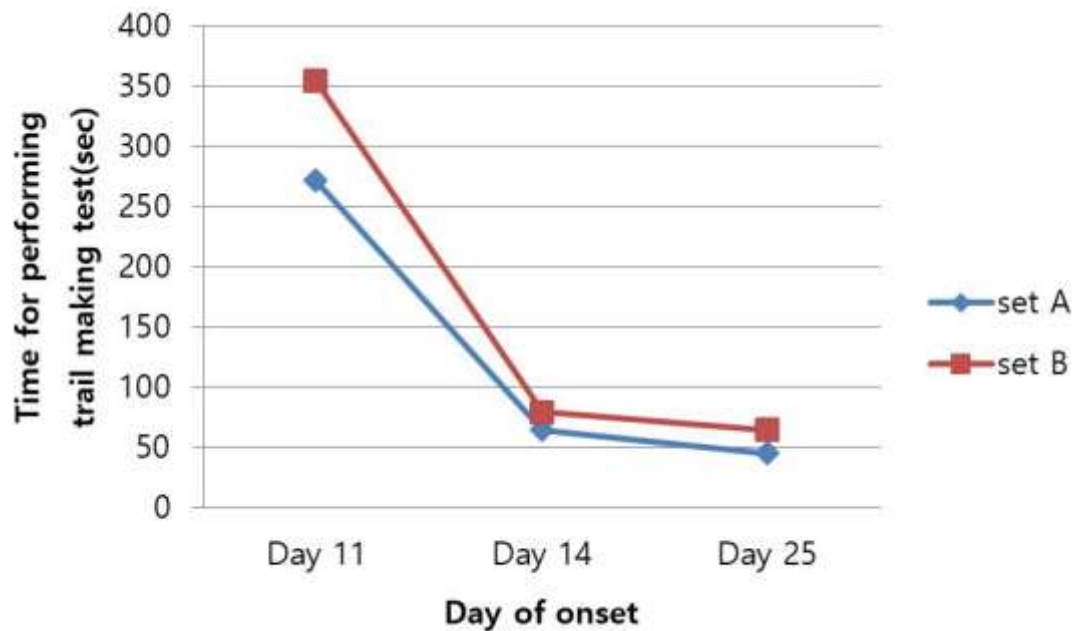


Fig 2. Trail making test for the evaluation of performance-based instrumental activities of daily living. Normative values of peers; set A 25.7 ± 8.8 ; set B 49.8 ± 15.2