—Case Reports—

Artery of Percheron Infarction with Bilateral Thalamic Lesions in a 14-Year-Old Girl: A Case Report

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The artery of Percheron (AOP), a common anatomic vascular variation of the P1 segment of the posterior cerebral artery, provides arterial blood supply to the paramedian thalami and rostral midbrain. Occlusion of the AOP can lead to infarction of the bilateral paramedian thalamus, with or without midbrain involvement, but is rare in children. Here, we describe a case involving a 14-year-old girl with sudden onset of disturbance of consciousness, hypersomnia, and global aphasia. Brain magnetic resonance imaging showed symmetric bilateral paramedian thalamic infarcts. Left-sided AOP infarction was diagnosed by brain angiography. (J Nippon Med Sch 2024; 91: 508–511)

Key words: artery of Percheron, infarction, children

Introduction

The artery of Percheron (AOP), a common anatomic vascular variation of the P1 segment of the posterior cerebral artery, is classified as type IIb according to the Percheron classification[1]. The AOP provides arterial blood supply to the paramedian thalami and rostral midbrain. Therefore, occlusion of the AOP can lead to an infarction of the bilateral paramedian thalamus, with or without midbrain involvement[2,3]. However, AOP occlusion is rare in children[4–7]. Here, we describe a case involving a 14-year-old girl who developed sudden onset of disturbance of consciousness, hypersomnia, and global aphasia. Brain magnetic resonance imaging (MRI) showed symmetric bilateral paramedian thalamic infarcts. Left-sided AOP infarction was diagnosed by brain angiography.

Case Report

A 14-year-old girl with difficulty awakening, even when stimulated, was admitted to the emergency department of our hospital. On arrival, her consciousness was impaired (Glasgow Coma Scale [GCS] score of 7 [E1V1M5]) and she had difficulty communicating; vital signs were normal. Laboratory evaluation of blood and cerebrospinal fluid, and the findings of brain CT, were unremarkable. On the sixth day of illness, her consciousness improved without treatment for ischemia, encephalitis, brain edema, or convulsions. She was discharged on the seventh day of illness. However, her consciousness remained impaired after discharge.

A few days after discharge, prolonged consciousness disorder reappeared along with hypersomnia and global aphasia, and she was referred to our pediatrics department. Electroencephalography (EEG) examination showed continuous bilateral high-amplitude slow waves without seizure discharge (**Fig. 1a**). She was admitted to the pediatric department on the 15th day after the onset of symptoms.

She was born at full term after a normal pregnancy and delivery. Her family history and past medical history revealed no evidence of conditions such as hypercoagulation syndrome, congenital heart disease, congenital metabolic disease, carbon monoxide intoxication, alcohol consumption, or medication use.

On admission, her temperature was 37.0°C, heart rate 52 bpm, and blood pressure 114/68 mm Hg. She exhibited sudden onset of disturbance of consciousness and speech difficulties. Her GCS score was 8 (E2V2M4). Pain stimuli caused eye-opening and escape reaction, with un-

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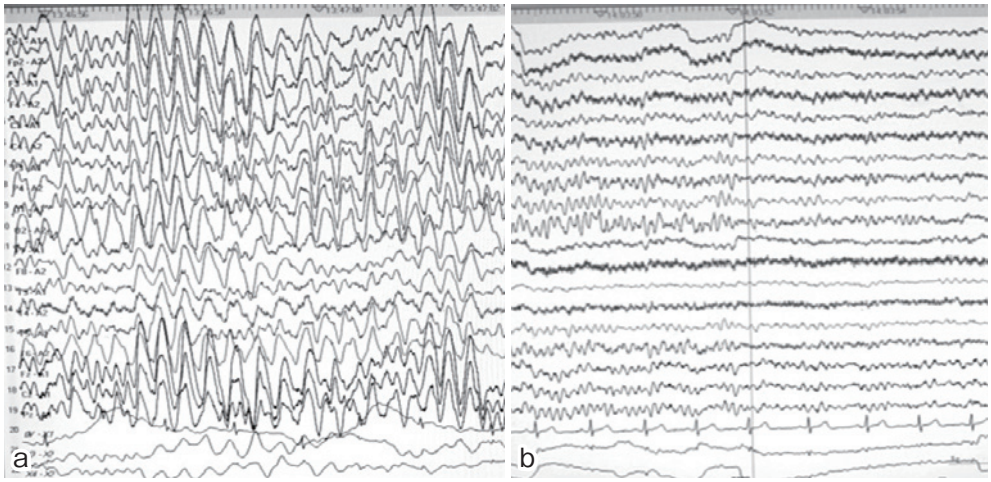

Fig. 1
a: An electroencephalogram on the 15th day after onset of symptoms showed continuous bilateral high-amplitude slow waves without seizure discharge.
b: Electroencephalography findings were normal on the 43rd day after onset of symptoms.

intelligible vocalizations. She did not exhibit meningeal irritation. Pupil size was 2.5 mm/2.5 mm (right/left) with a prompt light reflex. Facial perception was unknown. Eye movement showed no nystagmus, but fixing was difficult. Deep tendon reflexes, including the Achilles and patellar tendon, were attenuated on both sides, and the biceps tendon reflex was observed only on the left side. She did not show pathological reflex or involuntary movements. We suspected that tendon reflexes were increased. The discrepancy is likely attributable to difficulty in conducting a neurological examination of a patient with an acute illness. A manual muscle strength test showed a score of 1-2, although her consciousness disorder persisted for nearly 1 month. We suspected that muscle strength might have declined; however, assessment was challenging because voluntary movement is difficult to evaluate. She did not report thalamic pain.

Transesophageal and chest wall echocardiography showed normal cardinal construction and no intracardiac thrombosis. The findings of an electrocardiogram (ECG) and Holter ECG were normal. Doppler ultrasound showed no evidence of deep venous thrombosis in the cervical or lower extremity arteries. Prothrombin times and partial thromboplastin times were normal. Coagulation factors II, V, anti-thrombin III, protein C, protein S, and anti-cardiolipin antibodies, which cause hypercoagulation syndrome, were all within normal limits. Serum homocysteine and vitamin B_1 levels, alpha-galactosidase activity, ammonia, lactate, pyruvate, and blood gas analysis findings were normal. Screening tests for organic and amino acid metabolism were normal. Cerebrospinal fluid examination showed normal cell counts, lactate, pyruvate, and myelin basic protein, with 51 mg/dL of total protein.

Because we suspected that her condition involved autoimmune and herpes encephalitis, she was treated with high-dose gamma globulin therapy, steroids, edaravone, and acyclovir. Brain MRI on the 19th day of illness revealed hyperintense bilateral symmetric lesions in the thalamus on T2-weighted and fluid-attenuated inversion recovery images (**Fig. 2**). However, magnetic resonance angiography showed no blood vessel abnormalities.

MRI findings suggested acute necrotizing encephalopathy, so steroid pulse therapy with heparin was started. Cerebral blood flow scintigraphy (20th day of illness) showed no obvious local decrease in blood flow, and no abnormalities were seen on magnetic resonance venography (26th day of illness). However, three-dimensional CT angiography on the 28th day of illness could not identify the thalamic perforating artery (**Fig. 3**). Because of the possibility of thalamic perforating artery infarction, heparin treatment (7,500 U/day) was continued.

On the 43rd day of illness, EEG findings were normal (**Fig. 1b**). On the 67th day of illness, cerebral angiography was performed. A thalamic perforating artery branching from the anterior posterior cerebral artery (P1) was not observed (**Fig. 4**). No findings suggested dissection, stenosis irregularity, or vasculitis in the aortic arch, basilar artery, or posterior cerebral artery.

Ultimately, cerebral angiography findings and her clinical course indicated a diagnosis of AOP infarction. Because we could not identify the underlying disease

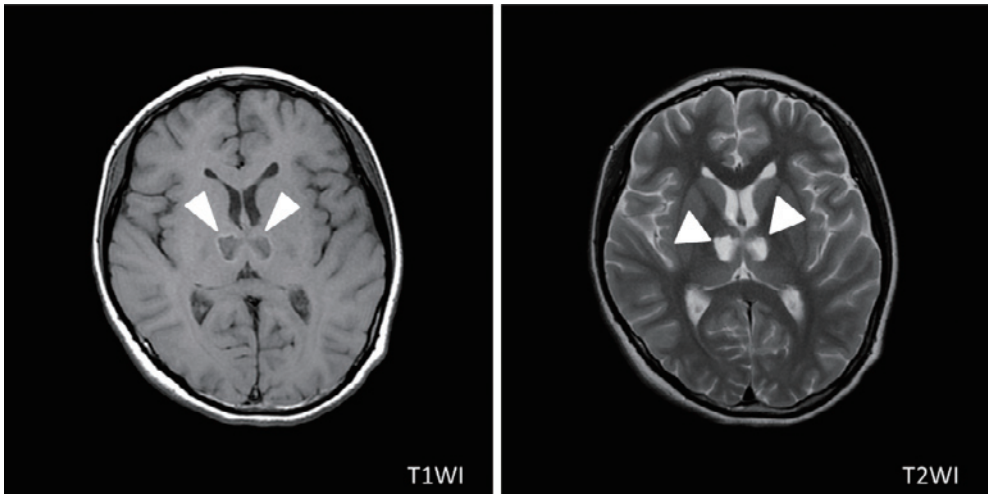

Fig. 2 Brain MRI on the 19th day of illness showed bilateral symmetric lesions in the thalamus that were hyperintense on T2-weighted and fluid-attenuated inversion recovery images (arrowheads).

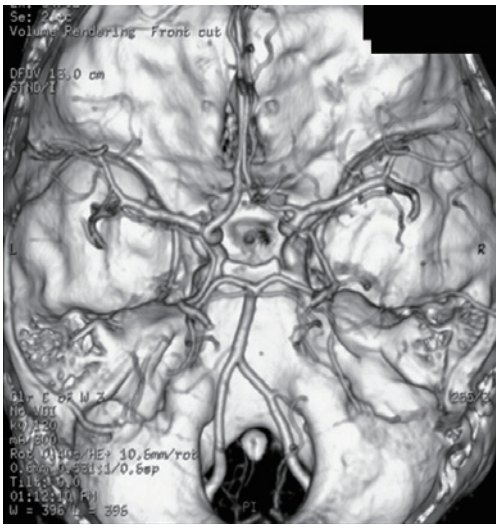

Fig. 3 No thalamic perforating artery could be identified on a three-dimensional CT angiogram on the 28th day of illness.

that had caused the stroke, she was maintained on aspirin treatment after heparin administration.

Her hypersomnia and prolonged disturbance of consciousness gradually improved within a few months. Rehabilitation improved her cognitive and motor function, and she has recently regained the ability to speak and ride a bicycle.

Discussion

The thalamic arteries consist of a group of perforating arteries of the thalamus that originate from the posterior communicating and cerebral arteries, including the thalamogeniculate and medial and lateral posterior choroidal arteries. The symptoms of thalamic perforating artery infarction are disturbance of consciousness, neuropsychiatric symptoms, aphasia, memory impairment, and vertical ocular motility disorder. Daytime hypersomnia may also be present. Hypersomnia symptoms improve within a few months, but sleep quality may remain impaired. In the chronic phase, sequelae such as higher dysfunction and personality changes are seen. To classify this infarction, we performed various examinations, including testing of coagulation and fibrinolysis function, cardiac/lower extremity/cervical ultrasonography, and infectious disease antibody titers. However, the findings were unremarkable, as well as her family history. We could not identify the cause of infarction, although lacunar infarction, atherothrombosis, and cardiogenic cerebral embolism were considered. We therefore classified her infarction as due to other/unidentifiable causes.

The differential diagnosis for disorders caused by bilateral thalamic lesions include 1) vascular diseases such as embolic territorial infarction, chronic hypertensive encephalopathy, and deep cerebral venous thrombosis; 2) congenital metabolic diseases such as Leigh's syndrome, GM2 gangliosidosis, Krabbe disease, and Wilson's disease; 3) acute necrotizing encephalopathy; 4) infectious diseases; 5) acquired metabolic and degenerative diseases such as Creutzfeldt-Jakob disease, Wernicke encephalopathy, Fahr's disease, osmotic demyelination syndrome, and profound hypoxia in newborns; 6) tumors such as gliomas, germinomas, and teratomas; and 7) demyelinating disorders such as multiple sclerosis and acute demyelinating encephalomyelitis[6]. The present findings highlight the need to consider AOP infarction in patients with bilateral thalamic lesions, even children, and provide in-

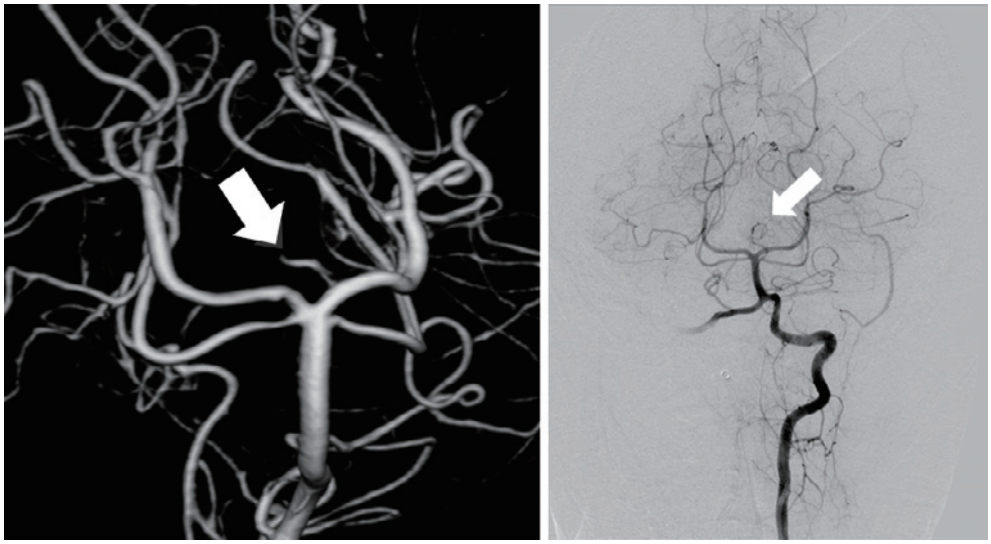

Fig. 4 A thalamic perforating artery branching from the anterior posterior cerebral artery (P1) was not visible on cerebral angiograms on the 67th day of illness. The arrows show the superior cerebellar artery (SCA).

sight on treatment decisions related to antithrombotic and antiplatelet therapy when the underlying disease is unclear.

Conclusion

The findings for this case of AOP infarction in a 14-year-old girl indicate that AOP infarction should be considered in the differential diagnosis for bilateral thalamic lesions, even in children. In addition, like other cerebral infarctions, AOP infarction may not be evident on brain CTs obtained during the hyper-acute phase.

Conflict of Interest: None declared.

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