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CASE REPORT

A Rare Case with Artery of Percheron Infarction: A Mysterious Cause of Reversible Coma

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Herein, we illustrated a rare patient with artery of Percheron (AOP) infarction who presented as reversible coma. Through the presentation of this case and literature review, we point out the diagnostic challenges in this entity. Second, we discuss the possible mechanisms of recovery of consciousness in this stroke subtype and remark the need for future reports using advanced functional neuroimaging techniques to clarify the unknown mechanisms underlying the evolution of the clinic in AOP infarction. We believe that the results of these studies may also give crucial insights regarding the processing of some critical functions such as sleep, arousal, and consciousness as well as our understanding of various consciousness disorders.

Key words: Artery of Percheron, paramedian thalamic region, pathophysiology, reversible coma

INTRODUCTION

Artery of Percheron (AOP) is a rare anatomical variant describing a single thalamic perforating artery arising from the proximal posterior cerebral artery. This anatomical variant supplies the rostral mesencephalon and both paramedian thalami.[1] Proximal artery embolism is considered as the most common etiology.[1] AOP occlusion generally presents with symptoms including sleep-like coma, disorientation, confusion, and hypersomnolence.[2] Ocular movement disturbances may be involved in the clinic when infarct extends to the rostral midbrain.[2] Isolated sleep-like coma presentations in AOP occlusion, without a focal sign, may constitute a diagnostic challenge which may potentially cause a delay in diagnosis as well as prevent a satisfactory management.[3,4] In this report, we present a rare patient with AOP infarct at which the diagnosis was considerably hard. Based on the presentation of this case and literature review, we will discuss the underlying mechanisms of devastating spontaneous recovery of sleep-like coma in these clinical pictures.

CASE REPORT

A 58-year-old female patient presented to our emergency center after being found unresponsive by his relatives 4 h before admission. The last time she was seen normal was the previous night. On history interrogation, it was learned that the patient had had an ischemic stroke affecting the left parietal lobe 2 years ago presenting with mild right-sided hemiparesis which had totally recovered in the follow-up. However, the patient had not admitted to outpatient follow-ups, and she had stopped taking medicines a long time ago. On admission to the emergency center, the patient was noncooperative and nonorientated with a Glasgow Coma Scale (GCS) of 5 (E: 1, V: 1, and M: 3). Her pupils were 3 mm in diameter bilaterally and reactive to light. Not a focal neurological sign was found. The National Institutes of Health Stroke Scale score was 31 points, and the score of the modified Rankin Scale (mRS) was evaluated as 5 points. Remarkably, there was no history of intake of a toxic agent or a past medical history of a metabolic disturbance that could lead to the coma. The vital signs including blood pressure, heart rate, and body temperature were in the normal ranges. However, the patient had difficulty maintaining respiratory function due to severe coma. The laboratory investigations including hemogram, full biochemistry, thyroid-stimulating hormone, B12, and folate were within the normal limits. Cranial computed tomography (CT) did not show hemorrhage and a new hypodensity other than left parietal encephalomalacia. Soon after the cranial CT, the patient was urgently intubated due to low GCS and apneic episodes. With provisional diagnoses of ischemic stroke and postictal state, phenytoin, aspirin, and atorvastatin treatments were initiated, and the patient was admitted to the intensive

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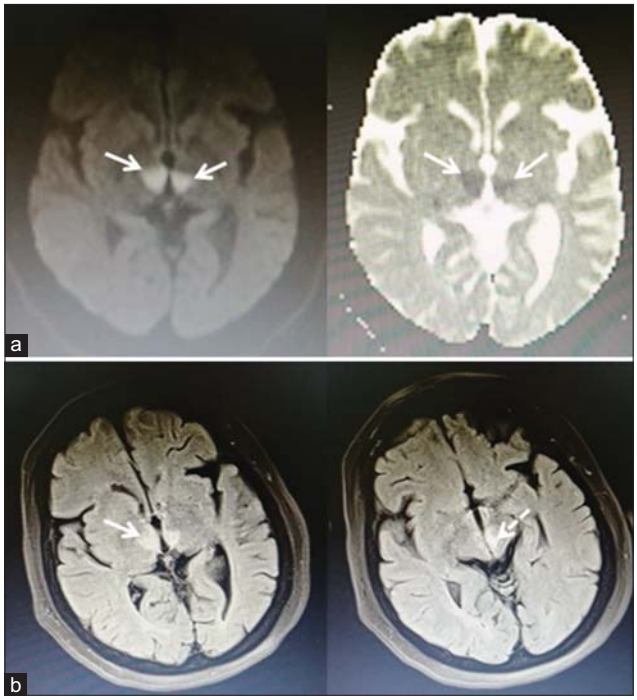

Figure 1: (a) Diffusion-weighted imaging and apparent diffusion coefficient sequences showing diffusion restriction in the bilateral paramedian thalamic region (arrows). (b) Images showing hyperintensity on fluid-attenuated inversion recovery in the paramedian thalamic regions and rostral mesencephalon (arrows)

care unit. Cranial magnetic resonance imaging (MRI) was performed on the 2nd day of admission, which showed diffusion restriction in the bilateral paramedian thalamic and midbrain regions, yielding the diagnosis of AOP infarct [Figure 1]. In the follow-up, her awareness improved gradually up to GCS of 14 (E: 4, V: 4, and M: 6) on the 2nd day of admission; hence, she was extubated. Brain/neck tomography angiography showed normal proximal arteries. Echocardiography and 24-h rhythm monitoring (Holter) examinations were in the normal ranges. Further investigations for young stroke including mutation analyses and alpha-galactosidase were found to be in the normal ranges. Taken together, ischemic stroke secondary to intracranial atherosclerosis was diagnosed. Routine electroencephalography could be performed on the 4th day after stroke which revealed an 8-Hz parieto-occipital alpha-rhythm, and no an epileptiform discharge or ictal activity was recorded. Hence, phenytoin was stopped, and the patient was discharged on aspirin and atorvastatin therapies. On neurological examination at discharge, the patient was orientated and cooperative; however, she was evaluated as mild apathetic. Of note, a moderate fluctuation in her level of consciousness was apparent which persisted for the 1st week of hospitalization. At discharge, her relatives described a moderate deterioration in her short-term memory. Not a focal neurological deficit was apparent, and she could walk without support. A mild upgaze palsy was present. The score of mRS at discharge was 2 points.

DISCUSSION

AOP occlusion is a rare cause of coma in which recovery of the coma is the rule, although other abnormalities such as hypersomnia, attention deficit, and ocular motility disorders may persist in the long term.[5] Considering various causes resulting in coma, the diagnosis of active occlusion cancellation occlusion is dependent on neuroimaging studies, basically MRI. For recognition of this syndrome, MRI must be performed eventually. Although CT is more available and easy to perform, usually it fails to determine the lesion that potentially causes a delay in diagnosis as well as prevent satisfactory management such as thrombectomy therapy in patients admitting within the therapeutic window. However, there are several reports remarking the diagnostic challenges in this subtype of stroke,[6-9] such that many of these patients experience a delay in diagnosis and initiation of treatment, with the majority of diagnoses occurring outside the tissue plasminogen activator window.[8,9] Besides, an appropriate diagnosis is also important for a more accurate determination of the prognosis of the patient, especially while informing their relatives in the early period of the event. In this report, through the presentation of this index case, first, we point out the importance of recognition of this entity among clinicians. Other causes also might cause bilateral thalamic lesions such as exposure to toxic agents, some metabolic disturbances, or viral encephalitis which should be considered during making the differential diagnosis in this patient.[10,11] However, there was no fever or a clinical manifestation that might suggest encephalitis. Besides, the clinical onset was very abruptly excluding encephalitis. On the other hand, there was no history of exposure to a toxic agent, and a history of a metabolic disturbance was absent as well as the results of the laboratory investigations were in the normal ranges. Finally, the diffusion-weighted imaging (DWI) showed bilateral diffusion-restricted lesions of the thalamus and midbrain which precisely corresponded to the territory of AOP and, therefore, highly specific for AOP infarction.

However, another strictly crucial and rather underestimated issue is the impressive recovery of consciousness within a short time interval in AOP infarction. AOP infarction affects mainly the bilateral paramedian thalamic region, an injury of intralaminar thalamic nuclei (located in the paramedian thalamus), which plays an important role in arousal, and motivation is known to be responsible from apathy.[5] Thalamus and particularly the nuclei within the internal medullary lamina, together with the associated thalamocortical (TC) connections, have been indicated critically important for

human consciousness. Due to its critical localization and connections with multiple cortical regions, a small, localized lesion in the thalamus may result in a fully unconsciousness state, whereas a cortical lesion of a similar size may not lead to a significant clinical manifestation.[12] While various functional and structural inputs from thalamic reticular nuclei (TRNs) and TC nuclei to cortical targets act in this circuit, the cortical neurons in layer VI also send strong excitatory feedback back to both TRN and TC neurons, which completes the loop of TC circuit.[13] Besides, the TC circuit which consists of reciprocal connections between the thalamus and the cortex is known to function critically also in sleep-wake control.[13] Based on the current knowledge, we can explain the sleep-like coma presentation in our index case, similarly, by mechanisms of TC deafferentation, which was shown to occur during nonrapid eye movement sleep and negatively correlated with awareness state.[14] However, a much more interesting and unknown point is that by which mechanisms the brain reserve compensates this malfunction? In our patient, DWI showed bilateral diffusion restriction in the bilateral medial region of the thalamus (paramedian thalamic). Diffusion-restricted site reflects cytotoxic edema which is accepted as a sign of irreversible tissue damage. Hence, explaining the devastating rapid recovery of consciousness in this patient through structural recovery (or histopathological recovery) of thalamic nuclei would be probably irrational. In my opinion, the dramatic recovery of the patients with thalamic infarct can be hypothesized to be associated with alterations in the mentioned feedback from cortex to thalamic nuclei,[13] which probably compensates rapidly the functional impairment developing due to structural injury of the thalamus in the acute period. However, we can only hypothesize this view as advanced functional and structural neuroimaging methods to support these considerations were unavailable in our case.

Future reports of large case series using advanced functional neuroimaging techniques are warranted to clarify the unknown mechanisms underlying the evolution of the clinic. The results of these reports may also give crucial insights regarding the processing of some critical functions such as sleep, arousal, and consciousness as well as our understanding of various consciousness disorders.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understand that her name and initial will not be published and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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