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Transient coma as Percheron artery stroke A case report

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ABSTRACT

The artery of Percheron is a rare anatomic variation in the brain vascularization, in which a single arterial trunk arises from the posterior cerebral artery to supply both sides of brain structures, i.e., the thalamus and midbrain. Occlusion of this uncommon vessel results in a characteristic pattern of bilateral paramedian thalamic infarcts with or without mesencephalic infarctions. We report the case of a Caucasian woman who completely recovers after transient coma due to Percheron artery infarction.

KEY WORDS: Percheron's artery, Occlusion, transient coma, infarction

INTRODUCTION

The artery of Percheron (AOP) is a rare anatomic variation in the brain vascularization, in which a single arterial trunk arises from the posterior cerebral artery (PCA) to supply both sides of brain structures, i.e., the thalamus and midbrain. Occlusion of this uncommon vessel results in a characteristic pattern of bilateral paramedian thalamic infarcts with or without mesencephalic infarctions. We report the case of a Caucasian woman who completely recovers after transient coma due to Percheron artery infarction.

CASE REPORT

We report the case of a 76-year-old Caucasian woman who was admitted to the emergency room in coma state (Glasgow Coma Scale = 7). Her husband found her lying on the floor, unresponsive. Two hours before, she was in healthy conditions. Her history was unremarkable, except for arterial hypertension and a previous surgical intervention for the left popliteal aneurism. She has never had attacks of dizziness or loss of consciousness; she had never suffered from epilepsy. Neurological examination showed bilateral Babinski sign and bilateral myotic pupils with mild anisocoria (left > right), no gaze deviation, and snoring with pauses of 2-3 seconds. Brain computed tomography (CT), blood examinations, and electrocardiogram all performed in the emergency department were all normal. After an hour, her state of consciousness improved spontaneously and the patient became alert and cooperative. Her neurological examination became unremarkable except for mild bilateral limitation in upward gaze. Electroencephalography was normal.

Brain magnetic resonance imaging, performed after 24 hours from the beginning of the event, showed bilateral thalamic subacute stroke; occlusion of basilar apex was excluded (figure 1). The stroke of Percheron artery territory was diagnosed. At

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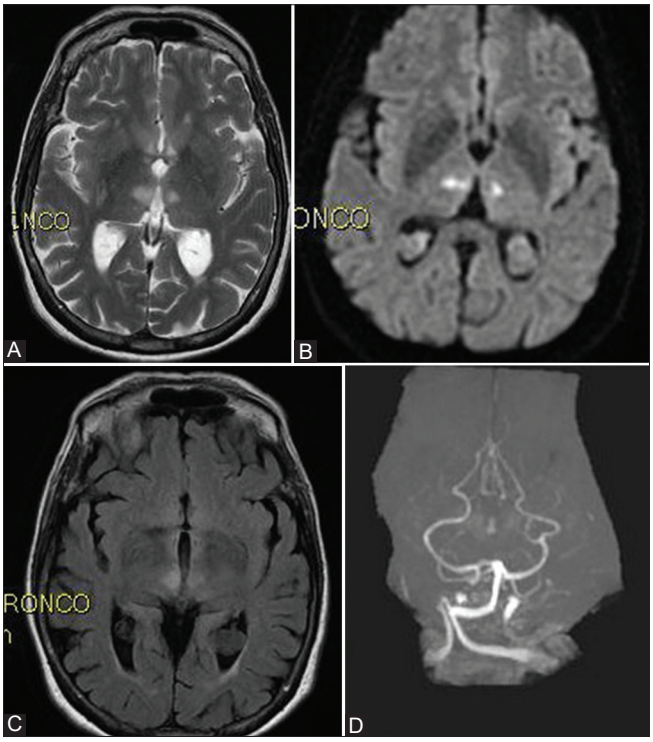

FIGURE 1. (A) T2 FRFSE sequence: Bilateral thalamic infarction. (B) Diffusion-weighted imaging sequence: Bilateral thalamic infarction. (C) T2 flair: Bilateral thalamic infarction. (D) Angio-magnetic resonance imaging: Normal basilar apex.

discharge, neurological examination and cognitive assessment were unremarkable.

DISCUSSION

The thalamus is predominantly supplied by multiple small vessels originating from the posterior communicating artery (PcomA) and P1 and P2 segments of the PCAs. Although there are significant variation and overlap, thalamic vascular supply is classically categorized into four territories: Anterior, paramedian, inferolateral, and posterior. The anterior territory is supplied by the polar (or thalamotuberal) arteries, which arise from the PcomA. The paramedian territory is supplied by the paramedian (or thalamoperforating) arteries, which arise from the P1 segment of the PCA. The inferolateral territory is supplied by the thalamogeniculate arteries, which arise from the P2 segment of the PCA. The posterior territory is supplied by the posterior choroidal arteries, which arise from the P2 segment of the PCA (1). In 1973, a French neurologist Gerard Percheron identified a rare single basilar artery branching variant, which supplies bilateral sides of paramedian thalamus. This rare variant artery was later named as the "AOP" or synchronous bilateral paramedian thalamic stroke (2). The AOP is a rare anatomic variant of arterial supply to the paramedian thalamus and rostral midbrain, and occlusion of the AOP results in bilateral paramedian thalamic infarcts with or without midbrain involvement (3).

Bilateral paramedian thalamic infarcts are rare (0.1-2% of ischemic strokes) (4). The main cause is the occlusion of the AP due to cardioembolism (5). Small vessel disease (33-38.9%); cardioembolic source (0-22%); large vessel disease (13.2-22.2%); other causes such as vasospasm secondary to subarachnoid hemorrhage, hemodynamic alterations, vascular dissection, distal ischemia of the PCA, aneurysms of the basilar artery, hypercoagulable states, and vasculitis secondary to infections of the central nervous system (13-15.7%); and idiopathic causes (10%) are other recognized risk factors for AOP (6).

Recent reviews have pointed out that the mean age of patients with this type of stroke ranges from 61 to 64 years, with a slight prevalence in males (7). Patients typically present with decreased level of consciousness, vertical gaze palsy, and behavioral and cognitive changes (akinetic mutism, confusion, drowsiness, hypersomnolence, or coma) (8). Patients with bilateral paramedian thalamic infarcts accompanied by rostral midbrain lesions also have hemiplegia, cerebellar ataxia, movement dysfunctions, and oculomotor deficits (3). Recently, a case report reported status epilepticus as the first presenting sign (9). We performed a brief review of literature with atypical presentations (Table 1).

Altered mental status presents on the spectrum from difficult arousal, confusion, and stupor to coma. Changed consciousness is understood as interruption of ascending reticular activating system that is localized in the mesencephalic neurons. These neurons project their fibers to both thalami and then to the cortex (10). Oculomotor and pupillary deficits are also implicated in mesencephalic lesions. Vertical gaze palsy is explained by disruption of supranuclear inputs that traverse the thalamus on their way to the rostral interstitial medial longitudinal fasciculus (11). It has been proved by existence of vertical gaze palsy in AOP cases without midbrain involvement (11). Memory defect is suggested to be a result from disruption of mammillothalamic tract and anterior thalami, both of which belong to the internal connected components of the Papez circuit (10).

These disorders of vigilance generally occur with sudden onset and may persist until death though cases of complete recovery have been documented (12) such happened to our patient.

Differential diagnoses include internal cerebral vein thrombosis, top of the basilar artery infarction, and

TABLE 1. Case reports on percheron artery stroke

Author	Journal	Sample population	Peculiar symptoms of presentation
Case reports			
Martin et al.	Int Med J 2018	(n=1)	Persistent diplopia
Morelli et al.	Neurol Sci 2018	(n=1)	Bilateral carotid artery dissection
Oliveira et al.	Sleep Sci. 2018	(n=1)	Excessive daytime sleepiness
Ince et al.	Neurocase. 2018	(n=1)	Persistent anterograde amnesia
Pitts-Tucker	BMJ Case Rep	(n=1)	Multiple episodes of transient loss of consciousness
Wei et al.	Medicine (Baltimore). 2017	(n=1)	Holmes tremor 7 months after
Kouassi et al.	Am J Case Rep. 2017	(n=1)	Acute-onset vascular dementia, HIV association
Sechler et al.	Neurology. 2017	(n=1)	Selective downgaze palsy
Niazi et al.	J Coll Physicians Surg Pak. 2017	(n=1)	Severe vertigo, diplopia, and ataxia
Pupi et al.	Clin Case Rep. 2017	(n=1)	Bilateral internuclear and internal ophthalmoplegia
Jun-Hyung Lee	J Clin Neurol. 2017	(n=1)	Severe sensorineural hearing loss in both ears
Osman	Clin Case Rep. 2017	(n=1)	Sleep-like coma without focal neurological signs
Caruso et al.	Vasc Health Risk Manag 2016	(n=1)	Top of the basilar syndrome
Vasconcellos et al.	Dement Neuropsychol. 2016	(n=1)	Thalamopeduncular syndrome
Lee et al.	Ann Rehabil Med. 2016	(n=1)	Acute pseudobulbar palsy
Zhou et al.	Case Rep Neurol Med. 2015	(n=1)	Korsakoff syndrome
Nalbantoglu et al.	Acta Neurol Belg 2016	(n=1)	Acute confusional state
Jain et al.	Eur J Paediatr Neurol. 2011	(n=1)	Five-year-old squint and head bobbing at the age of 8 months
Wang et al.	Case Rep Neurol Med. 2013	(n=1)	Seizure
Koutsouraki et al.	J Neurol Sci 2009	(n=1)	Hypophonia, memory dysfunction, time disorientation and apathy
García-Casares	Rev Neurol. 2008	(n=1)	Altered language, bradypsychia, right hemiparesis, and right hemisensory loss
Lin et al.	Acta Neurol Taiwan	(n=1)	Status epilepticus

infiltrating neoplasms (2). Moreover, Creutzfeldt–Jakob disease, Wernicke encephalopathy, and extrapontine myelinolysis should also be considered in differential diagnosis (13). On the other hand, bilateral medial thalamic lesions, radiographically demonstrated, must be differentiated from several different nonvascular pathologies, such as toxic processes (Wernicke's encephalopathy), metabolic alterations (extrapontine myelinolysis and Wilson's disease), acute necrotizing encephalopathy, neurodegenerative disease (dorsomedian thalamic nucleus involvement in Creutzfeldt–Jacob disease), infections (Epstein–Barr virus encephalopathy), and neoplastic forms (bilateral thalamic glioma) (14). Sometimes, demyelinating pathologies, such as acute disseminated encephalomyelitis, could be suspected (14).

The imaging modalities of choice for early diagnosis of AOP infarction are diffusion-weighted imaging (DWI) and fluid-attenuated inversion recovery (FLAIR) (2).

A previous study performed to characterize the complete imaging spectrum of AOP infarction reported four distinct patterns of AOP infarction: Bilateral paramedian thalamic with rostral midbrain (43%), bilateral paramedian thalamic without midbrain (38%), bilateral paramedian and anterior thalamic with midbrain (14%), and bilateral paramedian and anterior thalamic without midbrain (5%) (2). These four distinct patterns were consistent with known variations in the paramedian artery. Another distinctive imaging finding was a V-shaped hyperintense signal intensity on axial FLAIR and DWI images along the pial surface of the midbrain in the interpeduncular fossa, the sensitivity of which was about 67% in cases of AOP infarction with midbrain involvement (2).

The AOP is rarely visualized with conventional angiography and, to the best of our knowledge, only four authors have successfully demonstrated this variant 5, 6; it is too small to be visualized by CT angiography or magnetic resonance angiography (15).

The prognosis of thalamic infarctions is relatively good when compared to thalamic hemorrhages (16). This also applies to pediatric cases (17).

Few cases of AOP stroke responsible for life-threatening complications have been reported. This situation requires admission to the intensive care unit and involvement of a stroke unit team in the management decisions (18).

In a recent review of literature, the authors concluded that thrombolysis and intravenous heparin were effective first-line treatment options for emergent uncomplicated AOP occlusion followed by prescription of long-term anticoagulants, while non-emergent cases without midbrain involvement could be treated through rehabilitation and continual monitoring by medical staff (19).

CONCLUSION

We presented the case of 76 years old who completely recovered after Percheron artery infarction, whose first symptom was a transient coma state lasting about 1 hour. In our opinion in the emergency room, Percheron artery occlusion should be considered in patients with unexplained coma.

REFERENCES

1. Agarwal N, Chaudhari A, Hansberry DR, Prestigiacomo CJ. Redefining thalamic vascularization vicariously through Gerald Percheron: A historical vignette. World Neurosurg 2014;81(1):198-201.
https://doi.org/10.1016/j.wneu.2013.01.030.
2. Lazzaro NA, Wright B, Castillo M, Fischbein NJ, Glastonbury CM, Hildenbrand PG, et al. Artery of Percheron infarction: Imaging patterns and clinical spectrum. AJNR Am J Neuroradiol 2010;31(7):1283-9.
https://doi.org/10.3174/ajnr.a2044.
3. Sandvig A, Lundberg S, Neuwirth J. Artery of Percheron infarction: A case report. J Med Case Rep 2017;11(1):221.
https://doi.org/10.1186/s13256-017-1375-3.
4. Hawkes MA, Arena JE, Rollán C, Pujol-Lereis VA, Romero C, Ameriso SF. Bilateral Paramedian Thalamic Infarction. Neurologist 2015;20(5):89-92.
https://doi.org/10.1097/nrl.0000000000000047.
5. de la Cruz-Cosme C, Márquez-Martínez M, Aguilar-Cuevas R, Romero-Acebal M, Valdivielso-Felices P. Percheron artery syndrome: Variability in presentation and differential diagnosis. Rev Neurol 2011;53(4):193-200.
https://doi.org/10.33588/rn.5304.2011084.
6. Garcia-Grimshaw MA, Peschard-Franco M, Gutierrez-Manjarrez FA. Bilateral thalamic ischemic stroke secondary to occlusion of the artery of Percheron. Cureus 2018;10(5):e2676.
https://doi.org/10.7759/cureus.2676.
7. Rodriguez EG, Lee JA. Bilateral thalamic infarcts due to occlusion of the artery of Percheron and discussion of the differential diagnosis of bilateral thalamic lesions. J Radiol Case Rep 2013;7(7):7-14.
https://doi.org/10.3941/jrcr.v7i7.961.
8. Jiménez Caballero PE. Bilateral Paramedian thalamic artery infarcts: Report of 10 cases. J Stroke Cerebrovasc Dis 2010;19(4):283-9.
https://doi.org/10.1016/j.jstrokecerebrovasdis.2009.07.003.
9. Lin SF, Lin TC, Hu HH, Chen CI. Bilateral paramedian thalamic infarction presenting as status epilepticus: A case report and review of the literatures.

Acta Neurol Taiwan 2015;24(4):125-30.

10. Ropper MA, Klein JP. Adams and Victor's Principles of Neurology. 10th ed. United States: McGraw-Hill; 2014.
11. Clark JM, Albers GW. Vertical gaze palsies from medial thalamic infarctions without midbrain involvement. Stroke 1995;26(8):1467-70.

    https://doi.org/10.1161/01.str.26.8.1467.
12. Reilly M, Connolly S, Stack J, Martin EA, Hutchinson M. Bilateral Paramedian thalamic infarction: A distinct but poorly recognized stroke syndrome. Q J Med 1992;82(297):63-70.

    https://doi.org/10.1093/oxfordjournals.qjmed.a068650.
13. Lamboley JL, Le Moigne F, Have L, Tsouka G, Drouet A, Salamand P, et al. Artery of Percheron occlusion: Value of MRI. A review of six cases. J Radiol 2011;92:1113-21.

    https://doi.org/10.1016/j.jradio.2011.08.007.
14. Kamaşak T, Sahin S, Eyüboğlu İ, Reis GP, Cansu A. Bilateral Paramedian thalamic syndrome after infection. Pediatr Neurol 2015;52(2):235-8.

    https://doi.org/10.1016/j.pediatrneurol.2014.09.012.
15. Shea YF, Lin OY, Chang RS, Luk JK. Artery of Percheron infarction. Hong Kong Med J 2012;18:446.e1-2.
16. Schmahmann JD. Vascular syndromes of the thalamus. Stroke 2003;34(9):2264-78.
17. Garg BP, DeMyer WE. Ischemic thalamic infarction in children: Clinical presentation, etiology, and outcome. Pediatr Neurol 1995;13:46-9.

    https://doi.org/10.1016/0887-8994(95)00108-r.
18. Zappella N, Merceron S, Nifle C, Hilly-Ginoux J, Bruneel F, Troché G, et al. Artery of Percheron infarction as an unusual cause of coma: Three cases and literature review. Neurocrit Care 2014;20(3):494-501.

    https://doi.org/10.1007/s12028-014-9962-2.
19. Li X, Agarwal N, Hansberry DR, Prestigiacomo CJ, Gandhi CD. Contemporary therapeutic strategies for occlusion of the artery of Percheron: A review of the literature. J Neurointerv Surg 2015;7(2):95-8.

    https://doi.org/10.1136/neurintsurg-2013-010913.