CASE REPORT

Sleep Science

A rare differential diagnosis of excessive daytime sleepiness - Artery of Percheron territory infarct

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ABSTRACT

Stroke is an example of neurological diseases that can commonly drives Excessive Daytime Sleepiness (EDS). Extensive strokes with brain edema can leave a brain herniation and coma. Other causes of EDS after stroke are strategic lesions at Thalamus and brainstein. A 56-year-old man, right handed, with hypertension and hypercholesterolemia was admitted at Emergency Room due to 5 days onset of EDS, memory impairment, and left-sided weakness. A brain magnetic resonance imaging showed paramedian thalamic hyperintensity with rostral midbrain hyperintensity extending along the pial surface of the interpeduncular fossa. The artery of Percheron (AP) is an unusual anatomical variation that originates from the posterior cerebral artery and irrigates the paramedian regions of the thalamus and part of the midbrain. It is important the clinical suspicions with detailed drowsy patients history. Awareness of the clinical and neuroimaging features of this stroke syndrome is essential for timely diagnosis and appropriate management.

Keywords: Stroke; Sleep; Midline Thalamic Nuclei; Midbrain Reticular Formation.

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INTRODUCTION

We present a 56-year-old man who was admitted at Emergency Room of University Hospital of Universidade Federal de São Paulo with initial complaint of excessive daytime sleepiness (EDS), left-sided weakness and sudden onset memory loss beginning 5 days ago. Symptoms started around 2pm, when he was found by his wife in the bedroom, drowsy and unable to get up. He was right handed and previously diagnosed with arterial hypertension and hypercholesterolemia, regularly using losartan 50mg/day and simvastatin 20mg/day. There was no history of daytime sleepiness before this period. There was also no history of fever, headache, intoxication, head manipulation, trauma or neck pain.

At hospital admission, his blood pressure was 135x85mmHg and serum glucose was 98mg/dl. Neurological examination showed a sleepy patient with Epworth Sleepiness Scale (ESS) of 14, dysarthria, motor function leftsided with grade 2/5 in upper extremities and 3/5 in lower extremities, and hypoesthesia in right side of body. Deep tendon reflexes on the left side were +3/4+ and a Babinski signal was found on the same side. After that, he was immediately hospitalized for clinical treatment and complementary investigation.

A brain computed tomography (CT) scan was performed one and a half hour after admission and showed a right thalamocapsular region infarct. Brain magnetic resonance imaging (MRI) was performed the day after the admission showing paramedian thalamic hyperintensity (Figure 1) and rostral midbrain hyperintensity extending along the pial surface of the midbrain of the interpeduncular fossa, characterizing the "V" sign (Figure 2) on FLAIR and T2-weighted images.

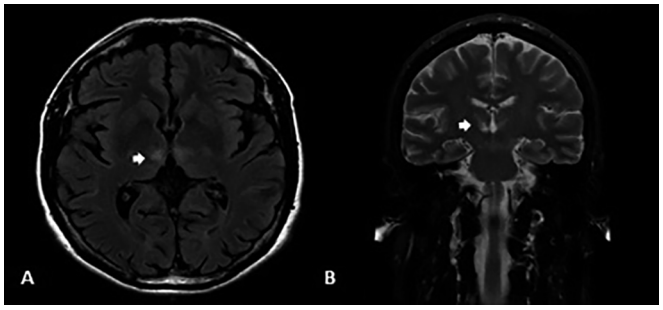

Figure 1. Axial FLAIR (A) and coronal T2-weighted (B) brain MRI showing with paramedian thalamic hyperintensity (arrows).

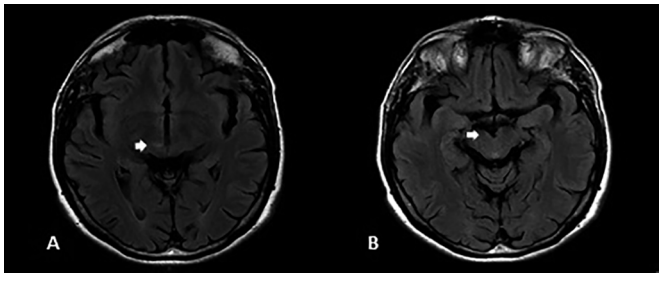

Figure 2. Axial FLAIR brain MRI showing rostral midbrain infarct (A) and a V-shaped hyperintense signal intensity along the pial surface of the midbrain at the interpeduncular fossa (the 'V' sign, indicated by arrow).

Currently, 4 weeks after symptoms onset, patient is accompanied at neurology clinic and does rehabilitation with physio and speech therapies. He presented improvement of motor function (grade 4/5 on both upper and lower left extremities) and EDS (ESS=8).

DISCUSSION

There are many causes of EDS. Sleep deprivation, sleep disorders, psychiatric diseases, use of medications, circadian rhythm sleep-wake disorders, and neurological diseases are examples of differential diagnosis. Stroke is an example of neurological diseases that can commonly drives EDS, with high prevalence and enourmous impact in our society[1].

The overlap among stroke and sleep diseases is an authentic issue. Stroke patients have higher prevalence of sleep diseases such obstructive sleep apnea, restless leg syndrome, and others. Unfortunately, the usual scales to identify the EDS in this population are not useful. Systematic investigation can be done to identify sleep problems[2].

On the other hand, acute lesions of CNS can develop EDS. Extensive strokes with brain edema can leave a brain herniation and coma. Other causes of EDS after stroke are strategic lesions at thalamus and brainstein[3]. Artery of Percheron (AP) is an unusual anatomical variation that originates from the posterior cerebral artery and irrigates the paramedian regions of the thalamus and part of the midbrain[4]. When this artery is occluded, usually by cardioembolic cause, there is a characteristic neuroimaging pattern: bilateral paramedian thalamic infarctions with or without involvement of midbrain[5,6].

Interestingly, the obstruction of the AP cannot usually be seen by angiography. It is important clinical suspicions with detailed drowsy patient's history. Awareness of the clinical and neuroimaging features of this stroke syndrome is essential for timely diagnosis and appropriate management.

Patients with AP infarct classically have sleepiness, vertical gaze paresis and memory impairment[6]. Sleep/wake disturbances were observed in patients with AOP infarction from stroke onset or become evident following recovery from coma, however, a few clinical presentations were described having EDS as initial complaint[3,7,8]. Altered mental status can present anywhere on the spectrum from drowsiness or confusion to hypersomnolence or coma[6,8]. These disorders of vigilance generally occur with sudden onset and may persist until death, though cases of complete recovery have been documented[9,10]. In our patient the EDS was very imperative as initial complaint improving after few weeks.

Paramedian thalamic stroke due to AP infarct is a cause of organic hypersomnia, which in the absence of systematic sleep-wake studies has been attributed to disruption of ascending activating impulses and considered a "dearoused" state[11]. Bassetti et al.[7] showed that paramedian thalamic stroke is accompanied not only by a dearousal but also a disruption of NREM sleep, which parallels the severity of hypersomnia. Hypocretin deficiency was not involved in hypersomnia observed in patients after AP infarction[12].

Further studies are needed to explain pathophysiological mechanisms and therapeutic possibilities involving patients with EDS following AP infarction, as well as factors which contribute for their outcomes.

REFERENCES

1. Mello-Fujita L, Kim LJ, Palombini Lde O, Rizzi C, Tufik S, Andersen ML, et al. Treatment of obstructive sleep apnea syndrome associated with stroke. Sleep Med. 2015;16(6):691-6. DOI: http://dx.doi.org/10.1016/j.sleep.2014.12.017
2. Arzt M, Young T, Peppard PE, Finn L, Ryan CM, Bayley M, et al. Dissociation of obstructive sleep apnea from hypersomnolence and obesity in patients with stroke. Stroke. 2010;41(3):e129-34. DOI: http://dx.doi.org/10.1161/STROKEAHA.109.566463
3. Hammersley D, Arora A, Dissanayake M, Sengupta N. Fluctuating drowsiness following cardiac catheterisation: artery of Percheron ischaemic stroke causing bilateral thalamic infarcts. BMJ Case Rep. 2017;2017. pii: bcr2016218035. DOI: http://dx.doi.org/10.1136/bcr-2016-218035
4. Almamun M, Suman A, Arshad S, Kumar SJ. A Case of Midbrain and Thalamic Infarction Involving Artery of Percheron. J Clin Med. 2015;4(3):369-74. DOI: http://dx.doi.org/10.3390/jcm4030369
5. 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. DOI: http://dx.doi.org/10.1007/s12028-014-9962-2
6. 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. DOI: http://dx.doi.org/10.3174/ajnr.A2044
7. Bassetti C, Mathis J, Gugger M, Lovblad KO, Hess CW. Hypersomnia following paramedian thalamic stroke: a report of 12 patients. Ann Neurol. 1996;39(4):471-80. PMID: 8619525 DOI: http://dx.doi.org/10.1002/ana.410390409
8. Hermann DM, Siccoli M, Brugger P, Wachter K, Mathis J, Achermann P, et al. Evolution of neurological, neuropsychological and sleep-wake disturbances after paramedian thalamic stroke. Stroke. 2008;39(1):62-8. DOI: http://dx.doi.org/10.1161/STROKEAHA.107.494955
9. Khoiny A, Goldberg M, Khoiny N. Atypical presentation with good outcome in a bilateral paramedian thalamic infarction. J Neurol Sci. 2006;23(1):54-8.
10. Krolak-Salmon P, Croisile B, Houzard C, Setiey A, Girard-Madoux P, Vighetto A. Total recovery after bilateral paramedian thalamic infarct. Eur Neurol. 2000;44(4):216-8. PMID: 11096220 DOI: http://dx.doi.org/10.1159/000008239
11. Guilleminault C, Quera-Salva MA, Goldberg MP. Pseudo-hypersomnia and pre-sleep behaviour with bilateral paramedian thalamic lesions. Brain. 1993;116(Pt 6):1549-63. DOI: http://dx.doi.org/10.1093/brain/116.6.1549
12. Suzuki K, Miyamoto T, Miyamoto M, Maeda H, Nokura K, Tohyama J, et al. Hypocretin-1 levels in the cerebrospinal fluid of patients with Percheron artery infarction with or without midbrain involvement: A case series. Medicine (Baltimore). 2016;95(29):e4281. DOI: http://dx.doi.org/10.1097/MD.0000000000004281