REVISTA NEUROCIÊNCIAS

Relato de Caso

Artery of Percheron territory infarct: an unusual cause of excessive daytime sleepiness

Infarto em território da artéria de Percheron: uma causa incomum de sonolência excessiva diurna

Infarto en el territorio de la arteria de Percheron: una causa infrecuente de somnolencia diurna excesiva

Vinícius Lopes Braga[1], Pedro Henrique Almeida Fraiman[2], Fernando Morgadinho Santos Coelho[3]

1.Discipline of Neurology, Department of Neurology and Neurosurgery, Escola Paulista de Medicina, Universidade Federal de São Paulo. São Paulo-SP, Brazil. Orcid: https://orcid.org/0000-0001-6736-2928
2.Discipline of Neurology, Department of Neurology and Neurosurgery, Escola Paulista de Medicina, Universidade Federal de São Paulo. Hospital Universitário Onofre Lopes, Division of Neurology, Universidade Federal do Rio Grande do Norte. Natal-RN, Brazil. Orcid: https://orcid.org/0000-0002-5250-8602
3.Discipline of Neurology, Department of Neurology and Neurosurgery and Discipline of Psychobiology, Escola Paulista de Medicina, Universidade Federal de São Paulo. São Paulo-SP, Brazil. Orcid: https://orcid.org/0000-0002-8482-3754

Resumo

Uma paciente de 23 anos é admitida com queixa de sonolência excessiva diurna (SED) e um episódio de alucinação visual há 3 dias. Após isso, sonolência intensa e ataques de sono. O exame neurológico era normal, exceto pela presença de sonolência. A ressonância magnética mostrou lesão talâmica paramediana bilateral. O conjunto dos achados confirmou o diagnóstico de infarto no território da Artéria de Percheron. SED é uma queixa neurológica comum e abrange um enorme espectro de diagnósticos diferenciais. Nos relatamos um caso raro de SED devido a um acidente vascular isquêmico em tálamo bilateral.

Unitermos. Acidente vascular cerebral; Sono; Artéria de Percheron; Tálamo; Sonolência excessiva diurna

Abstract

A 23yo female comes to the emergency department complaining of excessive daytime sleepiness (EDS) and one episode of visual hallucination three days ago. After that, permanent mild dizziness, and sleep attacks. The neurology exam was normal, except by subjectively drowsy but aroused easily to voice. MRI shows symmetrical hyperintense lesions on the paramedial Thalamus. The findings above confirmed the diagnosis of an acute infarction of the artery of Percheron. EDS is a common complaint in neurologic practice and has a wide spectrum of differential diagnoses. We describe an unusual cause of EDS caused by a stroke in the bilateral Thalamus.

Keywords. Stroke; Sleep; Artery of Percheron; Thalamus; Excessive daytime sleepiness

Resumen

Una mujer de 23 años acude al servicio de urgencias quejándose de somnolencia diurna excesiva (SDE) y un episodio de alucinación visual hace tres días. Después de eso, mareos leves permanentes y ataques de sueño. El examen de neurología fue normal, salvo por subjetivamente somnoliento pero que se despertaba con facilidad a la voz. La resonancia magnética muestra lesiones hiperintensas simétricas en el tálamo paramedial. Los hallazgos anteriores confirmaron el diagnóstico de un infarto agudo de la arteria de Percheron. El SDE es una queja común en la práctica neurológica y tiene un amplio espectro de diagnósticos

diferenciales. Describimos una causa inusual de SDE causada por un accidente cerebrovascular en el tálamo bilateral.
Palabras clave. Accidente cerebrovascular isquémico; Dormir; Arteria de Percheron; tálamo; Somnolencia diurna excesiva

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Corresponding address: Vinícius Lopes Braga. Escola Paulista de Medicina, Universidade Federal de São Paulo, Department of Neurology e Neurosurgery. São Paulo-SP, Brazil. E-mail: viniciuslopesbraga@live.com

INTRODUCTION

Excessive daytime sleepiness (EDS) is a common complaint in neurologic practice. Many disorders can cause SED as sleep deprivation; sleep disorders: narcolepsy, circadian rhythm sleep-wake disorders, insomnia; psychiatric disorders: depression, anxiety, fatigue, and other neurologic disorders: Alzheimer's disease, Parkinson's disease, epilepsy, chronic pain syndromes, and stroke[1].

Sleep disorders are related to a higher risk of stroke and stroke can drive changes in sleep during acute and chronic aspects with insomnia and EDS, depending on each patient and affected territory[1]. Nonetheless, we aim to describe an unusual case of EDS caused by a stroke in the bilateral Thalamus.

METHOD

Cross-sectional, descriptive study, elaborated with data from the clinical history, physical examination, and complementary tests. For the development of the study, the rules and regulatory guidelines for research involving human beings number 150 of the National Health Council

(Brazil, 2015) were respected. The present study followed the CARE-writer guidelines. The present study was approved by the local ethics committee (CAAE number 09144819.1.0000.5292).

CASE REPORT

The patient was a twenty-three years old-female, single, Caucasian, college student. She is Christian catholic, and she was born and lives in São Paulo city. The patient comes to the Emergency Department complaining of excessive daytime sleepiness starting in the last three days. Before, the patient used to go to bed around 11 pm, sleep at 1:30 am, and wake up at 8:00 am. Currently, the patient sleeps around 18 to 20 hours per day. The patient refers to one episode of visual hallucination (sees worms in the trash) three days ago. After that, permanent mild dizziness, and sleep attacks. In a review of symptoms, the patient refers to brief sleep paralysis two years ago and no other complaints. Her Epworth sleepiness scale was fourteen of twenty-one. She doesn't know how to drive. She has a past medical history of depression and denied other comorbidities. She is allergic to diclofenac. She uses at-home escitalopram and a combined hormonal contraceptive. In social history, the patient was a non-smoker, occasional alcoholic beverage drinker, and no recreational drugs. No recent travel. No known environmental exposures. The patient denied significant family history. The vital signals were stable, and the

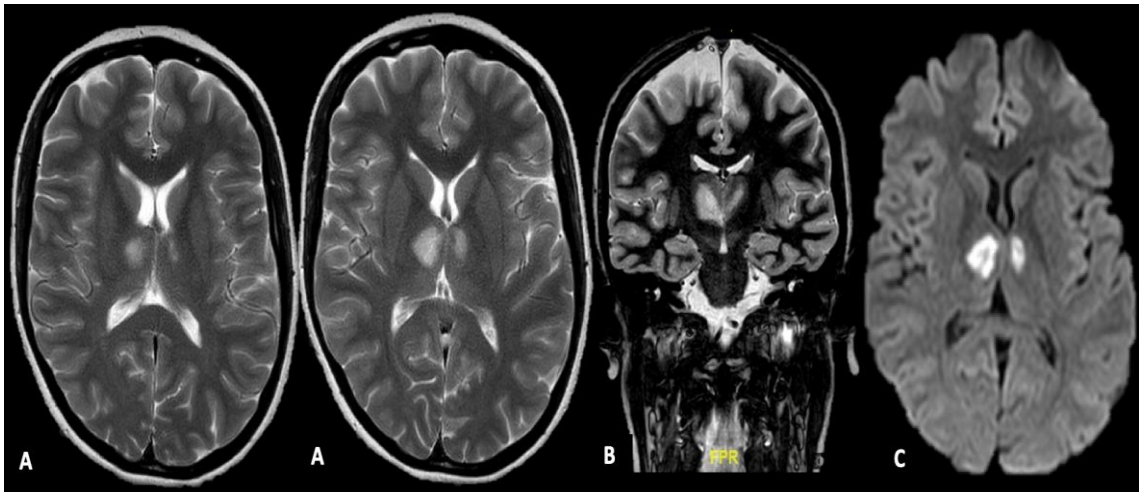
general exam was unremarkable. The neurology exam was normal, except by subjectively drowsy, but aroused easily to voice.

The general labs, cerebrospinal fluid, serum tests, rheumatological and hypercoagulability panel were normal. Axial FLAIR and T2-weighted MRI show symmetrical hyperintense lesions presented in the paramedial Thalamus with abnormal restriction of water diffusion on diffusion-weighted imaging, and a low apparent diffusion coefficient. (Figure 1) We prescribed aspirin and atorvastatin. The findings above confirmed the diagnosis of an acute infarction of the artery of Percheron. After that, we complete the investigation with an echocardiogram, 24-hour Holter, and micro-bubble transcranial doppler ultrasound. All three had normal results. To exclude the coexistence of narcolepsy, we performed a multiple sleep latency test, which shows no abnormalities. The findings above confirmed the diagnosis of an acute infarction of the artery of Percheron (AOP).

DISCUSSION

Thalamus is a large egg-shaped mass of grey matter situated at the core of the diencephalon and lies between the midbrain and forebrain, acting as a central hub. It amalgamates afferent and efferent axons traveling to and from the cerebral cortex.

Figure 1. **A**. Axial sections T2-weighted MRI show hyperintense lesions in bilateral Thalamus. **B**. Coronal section T2-weighted MRI shows hyperintense lesions in bilateral Thalamus. **C**. Restricted water diffusion on DWI.



Because of that, it is multifunctional and has a complex anatomy, recent papers say that the Thalamus consists of over a hundred nuclei[2]. The medial Thalamus is involved in memory and important for regulating arousal because it represents the superior extension of the midbrain reticular activating system. Infarction of the medial Thalamus can cause apathy and is a differential diagnosis from primary psychiatric disorders. The medial Thalamus is usually supplied by paramedian arteries. Bilateral lesions of the medial Thalamus can cause hypersomnia and decrease consciousness and include a wide spectrum of diagnoses[2-6] (Figure 2).

The artery of Percheron (AOP) is a rare anatomical variation presented in 4-12% of the population, first described by Gérard Percheron, when a single arterial trunk originated from the posterior cerebral artery irrigates the paramedian regions of the Thalamus. In some people, the

paramedian Thalamic arteries and the paramedian mesencephalic arteries to the brainstem originate from a common stem, the occlusion of which produces more extensive infarctions, accounting for midbrain and causing gaze palsies.

Figure 2. Differential diagnosis of bilateral Thalamic diseases.

Metabolic and toxic
• Wernicke encephalopathy • Osmotic demyelination syndrome • Fahr disease • Wilson disease • Fabry disease • Leigh disease • GM2 gangliosidosis • Krabbe disease
Primary neoplasm
• Glioma: grade II astrocytoma. • Primary CNS lymphoma • Paraneoplastic syndrome
Infection / Inflammatory Diseases
• Viral encephalitis • Creutzfeldt-Jacob disease • Vasculitis (Behçet) • Acute disseminated encephalomyelitis • Malaria, fungi, and toxoplasmosis
Vascular disorders
• Artery of Percheron infarct • "Top of the basilar" syndrome • Deep venous thrombosis • Hypotensive cerebral infarct • Posterior reversible encephalopathy syndrome

Bilateral paramedian Thalamic infarcts account for approximately 0.1% to 2% of all ischemic strokes and 27% of Thalamic infarcts. They are more frequent in men and the average age at presentation is around 60 years. Cardioembolic is the most common etiology. People with

AOP infarct have vertical gaze paresis (65%), neuropsychiatric symptoms, coma (42%), and memory impairment (58%). The disorders of awareness generally present from stroke onset in the spectrum that ranges from drowsiness or hypersomnolence to coma. In our patient, the EDS was the initial complaint and improved gradually after two months[2,6,7].

CONCLUSIONS

AOP's infarction are rare. It may cause diagnostic difficulties due to the variety of its clinical presentations and wide differentials, besides its small diameter and difficult visualization by diagnostic imaging – it can initially often be judged as normal. Clinicians must be aware of the possibility of AOP's infarction as a differential diagnosis of EDS, especially in the presence of sudden EDS onset or neuropsychological and ophthalmological signs.

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