Here we describe an elderly female patient with parkinsonism, symptomatic epilepsy and dementia due to multiple brain infarctions, associated to hemichorea-hemiballismus.

CASE REPORT

An 81-year-old female presented to the Neurology Service with sudden onset of involuntary random movements affecting her right hemibody characterized as hemichorea-hemiballismus. She had a past medical history of multiple ischemic strokes progressing to vascular dementia and parkinsonism, with irregular use of levodopa/benserazide 200/50 mg: ½ tablet tid. Additionally, she had a diagnosis of epilepsy with complex partial and secondary generalized seizures, using phenobarbital (100 mg/day) irregularly. On clinical examination, she had a severe cognitive impairment (Mini-Mental Status Examination 15/30) and right hemichorea-hemiballismus. During the bedside examination, she suffered an episode of complex partial seizures and, after that, the hemichorea-hemiballismus completely disappeared. After the complex partial seizure ended, hemichorea-hemiballismus re-emerged. She was managed with intravenous phenytoin (20 mg/kg) followed by maintenance treatment with oral valproic acid. She progressed with improvement of both her seizures and hemichorea-hemiballismus. A new reassessment was performed after 48 hours, and left upper limb rigidity, rest tremor and bradykinesia were documented, confirming the clinical diagnosis of parkinsonism probable due to vascular cause. A cranial computed tomography scan demonstrated multiple areas of brain infarctions, cortical atrophy and diffuse supratentorial hydrocephalus ex vacuo.

DISCUSSION

This case report illustrates the peculiar phenomenology of the association of MD that are traditionally viewed as mutually exclusive phenomena – a hypokinetic (parkinsonism) and a hyperkinetic (hemichorea-hemiballismus) disorder, associated with epileptic seizures, probably reflecting the involvement of multiple motor modulation pathways, in cortical and subcortical areas. The unpredictability of the clinical manifestations of the basal ganglia lesions, sometimes bordering on the inexplicable, was pointed out earlier as the Marsden's paradoxes: (1) different pathological lesions affecting similar sites might produce different clinical manifestations; (2) similar lesions impairing various basal ganglia might manifest with the same signs and symptoms; (3) the same lesions affecting the same areas produce a determined symptom or can be asymptomatic; and (4) certain disease affecting the basal ganglia have a wide range of MD[2]. In elderly patients, more susceptible to the occurrence of epilepsy, stroke is the most commonly associated risk factor, occurring in up to half of all cases with an identifiable etiology[3]. Hypokinetic and hyperkinetic MD can occur after an ischemic or hemorrhagic stroke in 1.0–3.7% of cases, notably hemichorea, hemiballismus and dystonia[4]. The neurological dysfunction of multiple domains in the same patient is a phenomenon that will probably accompany population aging and the increasing frequency of vascular events, polipharmacy and neurodegenerative diseases, among other factors[3-5]. The unusual case presented here represents a combination of diseases, each one with different underlying characteristics, and we named it as the chimera effect, as an allusion to Greek mythology.

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Bath-related headache: a Brazilian case report

Cefaleia relacionada ao banho: um relato de caso brasileiro

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The occurrence of headache triggered by bath (bath-related headache – BRH –) is rare. The first reference in the literature is of three middle-aged Japanese patients who, in 1992, developed symptoms of abrupt headache onset, of explosive and violent characteristic, caused by drop of hot water over the body in the shower which persisted for several weeks and resolved[1].

All the cases reported in the literature occurred in Asian populations[1,2], and there is a unique report on a Spanish woman[3]. We present a case of a Brazilian patient with no Asian ancestry.

Fifty-one-year-old woman, born and resident in Recife, Brazil, with no previous episodes of headache, had nicotine dependence and started varenicline, that was interrupted 30 days after two weeks of use.

The first episode occurred during the bath: sudden and very intense pain reaching the maximum power from the beginning. She described it as if something was to break the skull, and adjectives of "volcanic" and "cosmic" pain were expressed. She localized it on the "top of the head", bilaterally at the frontoparietal region. It lasted about 30 minutes slowly regressing, but persisting with mild intensity during the rest of the day. The response to analgesics was poor. Clinical exam was normal. MRI and arteriography ruled out the possibility of subarachnoid hemorrhage.

She evolved with daily crises, triggered by the water as it contacts the head. She began to avoid showering, fearing recurrence. Last episode occurred approximately 15 days after onset and, to date, she had no further crisis.

The contrast between the nature of an excruciating pain and the triggering of a so routine and trivial act is at least intriguing. The physicians themselves, facing this unusual situation and the picturesque character of the trigger, may tend to underestimate the condition and minimize it to a mere psychogenic conditioning factor.

It is assumed that BRH is a variant of idiopathic thunderclap headache, which may also have as a trigger a variety of factors, such as the Valsalva maneuver, sexual intercourse or strenuous exercise[4]. Etiopathogenic hypotheses involve excessive stimulation of temperature receptors in the skin and scalp and exaggerated autonomic neurovascular reflex, resulting in reversible cerebral vasoconstriction[4].

The pathogenic role of varenicline as a partial nicotinic cholinergic agonist is questioned. This is a new drug that includes in its side effects profile the onset of mild to moderate headache in 10.3%[5]. Although the symptoms occurred one month after it was discontinued, its participation cannot be completely rule out, since central nicotinic receptors have a role in brain vasoregulation.

The occurrence of BRH only in the Asian population was intriguing, but the report of this case and another one alike in this west part of the world[3] indicates that ethnic factors perhaps are not so decisive, and a universal occurrence would most likely exist. The disclosure of new cases may lead to a better nosological and etiopathogenic definition in the future.

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Medulla compression caused by vertebrobasilar dolichoectasia

Compressão medular causada por dolicoectasia vertebrobasilar

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