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Reversible cerebral vasoconstriction syndrome secondary to catheter ablation

Síndrome de vasoconstrição reversível secundária à ablação por cateter

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ABSTRACT

We report a case of a 41-year-old woman with chagasic cardiomyopathy who was submitted to ventricular septal catheter ablation. After the procedure she evolved with new-onset thunderclap headaches followed by migraine-type headaches. Cerebral angiography revealed bilateral segmental stenosis of the middle cerebral arteries.

Keywords: migraine, thunderclap headache, catheter ablation, Chagas' disease, tachyarrhythmia

RESUMO

Relatamos um caso de uma mulher de 41 anos de idade, portadora de cardiomiopatia chagásica, que foi submetida à ablação ventricular septal por cateter. Após o procedimento, a paciente apresentou cefaleia em trovoada de início recente seguida de cefaleia do tipo migranosa. A angiografia cerebral revelou estenoses segmentares bilaterais em território de artéria cerebral média.

Palavras-chave: migrânea, cefaleia em trovoada, ablação por cateter, doença de Chagas, taquiarritmia

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INTRODUCTION

The term thunderclap headache is used to describe an unanticipated severe headache reaching peak intensity within one minute. It is traditionally linked to aneurysmal subarachnoid hemorrhage but an increasing number of other causes have been identified. The reversible cerebral vasoconstriction syndrome (RCVS) is considered an underdiagnosed cause of thunderclap headache which predominanly affects middle-aged women and presents with reversible segmental cerebral vasoconstriction. Previously known as Call-Fleming syndrome or migraine angiitis, among other nomenclatures, the RCVS is commonly linked to post-partum period and vasoactive substances use. It frequently complicates with transient ischemic attacks, ischemic strokes, brain oedema, posterior reversible encephalopathy syndrome, and cerebral hemorrhages[1,2].

Chagas disease is endemic in Latin America and has become an emerging problem in developed countries because of international migrations[3]. The cardiac form is the most serious and frequent manifestations of chronic infection by the protozoan *Trypanossoma cruzi*. It typically leads to abnormalities of the conduction system, bradyarrhythymias and tachyarrhythmias, apical aneurysms, cardiac failure, thromboembolism, and sudden death[3].

We aim to describe the first case of a thunderclap headache followed by migraine-type headache in a patient who underwent ventricular septal radiofrequency catheter ablation (RCA) due to a severe chagasic cardiomyopathy.

CASE REPORT

A 41-year-old Brazilian woman with chronic cardiac Chagas disease became refractory to amiodarone therapy in October 2011 and was treated with implantable cardioverter defibrillator (ICD) to control ventricular tachyarrhythmic storms with haemodynamic compromise. At that time, echocardiography revealed a left apical ventricular aneurysm and left ventricular ejection fraction of 19%. In January 2012, she was readmitted due to cardiac shocks that telemetry proved to have been unleashed by discharges of her definitive ICD. A month later, she was submitted to ventricular septal RCA and her ICD was reprogrammed. During her intensive care unit (ICU) stay, a few hours after the cardiac procedure, the patient developed a thunderclap headache with no other neurological signs, but with a surge of systolic blood pressure, reaching 210 mmHg. After the excruciating bilateral occipital pain peaking at one minute subsided, the patient persisted with milder throbbing sensation associated with nausea, photophobia and phonophobia, and lasting around 17 hours. Thunderclap headache with this milder migraine-type pain recurred four times during her stay in the ICU. Her neurological examination was normal. Cranial computed tomography was unrevealing and non-traumatic cerebrospinal fluid analysis was clear with normal cytometry and biochemistry. Cerebral arteriography revealed bilateral segmental stenosis of the second and third middle cerebral arteries territories (Figure 1). Pulmonary oedema incidentally occurred during the angiographic study and prompted another ICU stay. Pain release was tempted with intravenous anti-inflammatory drugs and prophylaxis was started with nortriptyline titrated to 50 mg a day with partial response after two months. Of noteworthy mention on her previous history, the patient described only mild and episodic tension-type headaches. She was a non-smoker woman without drug-addiction or any other co-morbidity. She was under prescription of diuretics, beta-blockers and amiodarone. Adrenergic drugs, selective serotonin reuptake drugs or triptans had never been prescribed to the patient. In a four-month follow-up, the patient was not experiencing further episodes of migraine-type or thunderclap headaches.

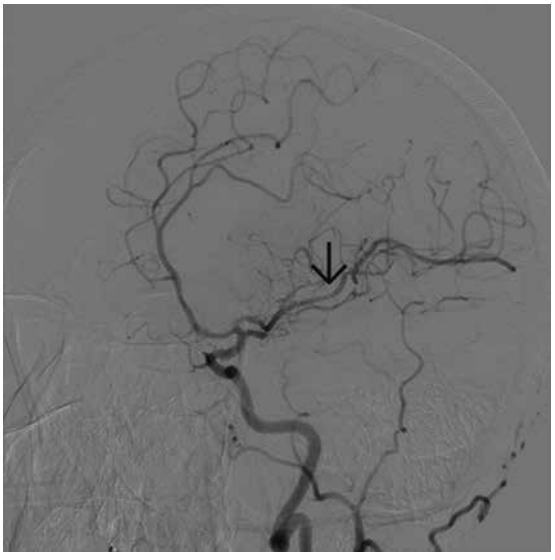

Figure 1. Cerebral arteriography – Arrow points to a segmental stenosis of the middle cerebral artery.

DISCUSSION

Herein reported is the first case of a thunderclap headache followed by migraine-type headache after a ventricular septal RCA to treat ventricular tachyarrhythmic storms in Chagas disease. Angiographic studies revealed cerebral vasoconstriction indicating the diagnosis of RCVS. New-onset migraine with or without aura have already been reported shortly after the RCA procedures for Wolff-Parkinson-White syndrome[4-6], atrial fibrillations[5-7] or other atrial septal defects[6,8].

RCVS is characterized by recurrent thunderclap headaches and reversible cerebral vasoconstrictions. RCVS is more common than previously thought and should mainly be differentiated from aneurismal subarachnoid hemorrhage, primary angiitis of the central nervous system and cerebral arterial dissections. RCVS's management and prognosis vary according to its cause. Treatment usually comprises the prescription of calcium-channel blockers due their supposedly effect on prevention of watershed ischaemia or cerebral hemorrhage[1,2].

Some may argue that thunderclap headache in RCVS may be a type of "crash migraine" since many patients have a previous history of migraine and the pain may present with typical characteristics of migraine. However, previous history of migraine is not an essential feature to the syndrome, as illustrated in our case. Besides, severe and abrupt onset is rather uncommon in migraine patients and angiographic studies during migraine crisis do not show cerebral vasoconstriction in the majority of cases[9].

Our patient developed a recurrent thuderclap headache followed by episodes of migraine-type headache associated with middle cerebral artery vasoespasm, typically present in RCVS. History suggests that ventricular septal RCA was the acute trigger event of the symptoms. The patient was satisfactorally treated with nortriptyline instead of calcium-channel blockers because of the latter's negative ionotropic effects which could harm the patient. The most specific finding of RCVS is the disappearance of cerebral vasoconstriction observed after 12 weeks in follow-up angiographic studies. A follow-up angiographic study was not performed in our patient because of her previous pulmonary complication after conventional angiography and the impossibility of performing magnetic resonance angiography due to the presence of ICD. Recent published analysis of a series of cases suggests that control angiography might not be necessary, since no differences in clinical features, frequency of laboratory abnormalities, CT/MRI findings, treatment, and clinical outcome between patients who underwent follow-up vascular imaging (angiography or transcranial Doppler ultrasonography) and those who did not[10].

This case report a new cause of RCVS among many increasingly recognized nowadays. We suggest that ventricular septal RCA should prompt a reflex mechanism of central pain sensitization mediated by peripheral vascular input, release of inflammatory mediators and subsequent cerebral vasoconstriction.

We declare no conflicts of interest/no financial support. Patient has consented with the publication of this manuscript.

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