

CASO CLÍNICO/CASE REPORT

Migraine with Aura After Endovascular Treatment of a Vertebro-Vertebral Fistula: Difficulties with Headache Classification**Enxaqueca com Aura Após o Tratamento Endovascular de Fístula Vertebro-Vertebral: Dificuldades na Classificação da Cefaleia**

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Abstract

Functional outcomes of endovascular treatment of arteriovenous fistulas have received little attention and it is unknown if the procedure may induce new headache.

We present a 16-year-old female, referred for observation due to complaints of headache, likely related to intracranial hypotension, after lumbar scoliosis correction surgery. Then she reported a discomfort in the right cervical region, where a central venous catheter had been previously placed. She performed Doppler and computed tomography angiography of the cervical vessels and a vertebra-vertebral fistula was identified on V1/V2 transition of the right vertebral artery. She underwent endovascular treatment, with successful fistula closure by coil embolization. Then, she started with new headache complaints, having suffered four episodes of headache similar to migraine with typical aura, which apparently resolved three months after the procedure.

We believe that headache with migraine characteristics could be considered secondary to endovascular treatment of arteriovenous fistula.

Resumo

Os resultados funcionais dos procedimentos endovasculares, nomeadamente o surgimento de cefaleia de novo, são pouco conhecidos.

Apresentamos o caso e uma adolescente de 16 anos, referenciada à consulta de Neuropediatria por queixas de cefaleia atribuída a hipotensão do líquido cefalorraquidiano, após uma cirurgia de correção de escoliose lombar. Posteriormente iniciou um desconforto na região cervical direita, onde tinha sido colocado um cateter venoso central. Realizou Doppler e angiografia por tomografia computadorizada dos vasos cervicais, tendo sido identificada uma fístula vertebro-vertebral na transição V1/V2. Realizou tratamento endovascular com o encerramento completo da fístula por embolização de coils, tendo iniciado posteriormente uma cefaleia com características de enxaqueca com aura típica, que resolveu três meses após o tratamento.

Consideramos que a cefaleia com características de enxaqueca pode ser secundária ao tratamento endovascular da fístula, embora não cumpra critérios de diagnóstico.

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Introduction

Vertebro-vertebral arteriovenous fistula (VV-AVF) is uncommon and is usually secondary to penetrating neck injury, blunt cervical trauma and iatrogenic forms of trauma, including direct percutaneous puncture of diagnostic angiography or during insertion of central venous catheters.¹ A surgical approach to VV-AVF is no longer warranted in most circumstances and the endovascular treatment (EVT) of the fistula has been increasingly applied and is currently considered the first therapeutic option, with a variety of devices available to fistula occlusion. However, functional outcomes after EVT, such as headache, have received little attention, and how a preexisting headache is affected by the procedure and the expected time frame to headache resolution are unknown clinical aspects. In fact, the few existing studies addressing these issues present conflicting results.^{2,3}

Patients who underwent intracranial endovascular procedures often develop headache in close temporal relation to the respective procedure. These patients often receive extensive diagnostic evaluations to determine the etiology of the headache, but the results of the testing usually provide little or no information that alters patient management or affects clinical outcome.^{4,5}

Theories about the genesis of headache associated with endovascular procedures include several aspects: mechanical stimulation of arterial wall by traction, with subsequent triggering of the trigeminovascular pathway, inflammatory changes caused by embolic or contrast materials, hemodynamic changes in collateral vessels or vascular spasm, physical and psychological stress, among others.

We present a patient that developed a headache with characteristics of migraine, after the endovascular treatment with coil embolization of a VV-AVF on V1/V2 transition.

Case Report

The patient, a 16-year-old girl, was referred to a Neurology consultation for complaints of bilateral frontal and occipital headache, described as a pressure (without any associated pulsatile quality), worsening with orthostatism and relieved when supine. She referred that this headache appeared immediately after the surgical treatment of lumbar scoliosis (apex in L1). The headache improved, with resolution in the first week after surgery, only with conservative measures. However, she started to experience discomfort in the right cervical region, where a central venous catheter had been placed, accompanied by a thrill. She was medicated with a daily

oral contraceptive (drospirenone + ethinyl estradiol) since she was 14-years-old and paracetamol (only when necessary). There was no personal history of headache or family history of migraine. The neurological examination was unremarkable.

She performed a Doppler of the cervical vessels and a cervical computed tomography angiography (angi-oCT) that suggested the presence of an arteriovenous fistula surrounding the right vertebral artery (**Fig. 1** and **2**). Digital subtraction angiography, by retrograde femoral catheterization, identified a right VV-AVF in V1/V2 transition (**Fig. 3**). She underwent EVT with coil em-

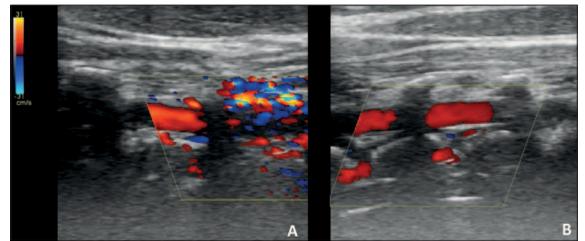


Figure 1. Right vertebral artery duplex ultrasonography. A: turbulent flow with high peak systolic velocity (> 300 cm/s) in V1/V2 transition; B: normal arterial flow distally to the V1/V2 transition.

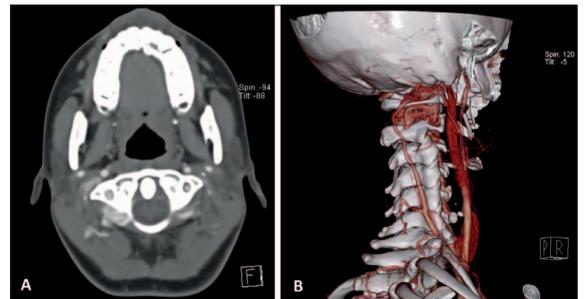


Figure 2. CT neck angiography. A: dilated vascular structures surrounding the right vertebral artery, probably related to arteriovenous fistula; B: three-dimensional reconstruction.

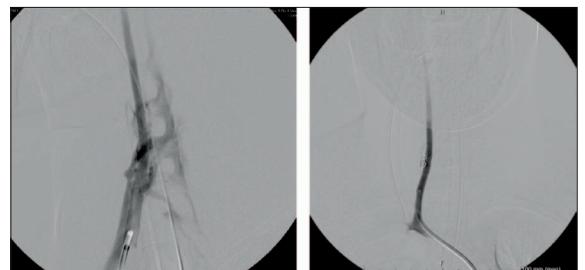


Figure 3. Cervicocerebral angiography. A: right vertebrovertebral fistula, with a fistulous tract at the V1/V2 transition; B: after endovascular treatment and fistulous tract closure with coils.

bolization with total exclusion of the fistula and no peri-procedure complications.

In the first week after the EVT, she experienced an episode of headache, with different characteristics from the previous ones, which recurred three times in a period of three months. All headache episodes were preceded by visual symptoms, described as binocular photopsias, without specified coloration, mobile and persisting with eyelid closure. While present, there was no modification of their intensity. The visual symptoms resolved within a few minutes, followed by a pulsatile, intense headache with bilateral frontal and occipital location, accompanied by nausea, vomiting, photo, phono and kinesiophobia. The pain, if untreated, lasted for several hours, but responded favorably to paracetamol. In one of the episodes, she also mentioned paresthesia in the lower limbs and spontaneous resolution in a few minutes, before the headache.

After these episodes, occurring in the first 3 months after the procedure, she did not present further events of this nature, remaining asymptomatic (having, so far, nine months of follow-up).

Discussion

This case highlights the complexity in the diagnosis of some headaches in pediatric age, being difficult to categorize them as primary or secondary. The patient initially presented (after lumbar scoliosis correction surgery) a condition that met criteria for the diagnosis of headache attributed to cerebrospinal fluid (CSF) hypotension, related to CSF fistula.⁶ Subsequently, an arteriovenous fistula involving the right vertebral artery was diagnosed in connection with orthopedic surgery (related to catheterization of a cervical vessel). The VV-AVF was successfully closed by endovascular procedure, with embolization with coils.

In the three months following fistula closure, she presented four episodes of headache that met the criteria for migraine with typical aura.⁶ The temporal relationship with angiography and EVT suggests, however, that the diagnosis could be of a secondary headache. In the presented case, the new headache complaints began one week after EVT with coil embolization and apparently resolved three months after the procedure. Three factors: (a) the location of the lesion (extracranial vertebral arteriovenous fistula); (b) type of endovascular procedure performed (coil embolization); and (c) time elapsed until the headache resolves (3 months) do not allow the classification as a secondary headache (attributed to an endarterial procedure), accordingly to the International Classification of Headache Disorders – 3rd edition (ICHD-III).⁶

The relationship with endovascular procedures has been studied by some authors, but the way in which headache is conditioned by the procedure and the time frame considered for this relationship are not well defined.^{2,4} Khan *et al* (2016) found that patients undergoing EVT of arteriovenous malformations and brain aneurysms had an increase in headache days three months later, compared to the previous month. In addition, a subset of patients was found to develop *de novo* migraine, which could persist for up to 2 years after the procedure. A new headache supports the causal relationship between EVT and headache.

So far, no studies trying to link EVT of extracranial vertebral arteriovenous fistula by coils embolization and new migraine-like headache complaints have been published (to our knowledge). As proposed by some authors, endovascular therapy can induce headaches by several mechanisms, such as local thrombosis, dilatation of the vessel wall and local inflammation after placement of coils.^{7,8} We can consider that the local release of inflammatory mediators may influence central pain modulation and can contribute to migraine-like attacks. We can also admit the eventual role in pain generation of regional changes in cerebral perfusion (due to adaptation to fistula closure) and/or the release of microemboli that can temporarily change the dynamic of cerebral circulation.⁹ Nevertheless, these are only speculative aspects that can be evoked trying to explain patient's complaints.

In conclusion, according to the criteria of ICHD-III, it is only possible to classify the headache of this patient as migraine with typical aura, because the occurrence of migraine attacks over a period of 3 months exceeds the time limit defined for a headache secondary to an endovascular intervention. Additional longitudinal studies may help to clarify the causality between these procedures and *de novo* headache complaints. ■

Responsabilidades Éticas

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