Hyperperfusion Syndrome After Flow Diverter Implantation for an Aneurysm of the Internal Carotid: a case report

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ABSTRACT

Treatment of intracranial aneurysms (IA), abnormal dilatations of a cerebral artery, can be performed either by surgical technique (clipping and/or wrapping) or endovascular technique (embolization or flow diverter implantation), the last one being based on the blood flow redirection and vascular remodeling, preventing blood from entering the aneurysm and resulting in thrombosis of the IA. In this case report, a rare complication of this procedure, the hyperperfusion syndrome, mostly seen in post-endarectomy procedures, and that has been poorly described in the literature is presented. Its mechanisms are not fully understood, though the main hypothesis includes endothelial lesion with loss of myogenic autoregulation of cerebral blood flow, resulting in symptoms such as seizures and focal neurological deficits that usually resolve in a few days.

Keywords: Hyperperfusion syndrome; Flow-diverting stent; Aneurysms

RESUMO

O tratamento dos aneurismas intracranianos (AI), dilatação anormal de uma artéria cerebral, pode ser realizado tanto por técnica cirúrgica (clipagem e/ou wrapping) quanto por técnica endovascular, com embolização ou implante de stent diversor/redirecionador de fluxo, sendo este último baseado em direcionamento do fluxo sanguíneo e remodelação do vaso, impedindo-o de entrar no aneurisma e resultando em trombose do AI. Neste relato de caso apresentamos uma complicação rara deste procedimento, o síndrome do hiperfluxo, mais observada em procedimentos pós-endarectomia, e que tem sido pouco descrita na literatura. Seus mecanismos não são totalmente compreendidos, sendo a principal hipótese a lesão endotelial com perda da autorregulação miogênica do fluxo sanguíneo cerebral, resultando em sintomas como convulsões e déficits neurológicos focais, que geralmente se resolvem em poucos dias.

Palavas-chave: Síndrome do hiperfluxo; Diversor de fluxo; Aneurisma

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INTRODUCTION

Intracranial aneurysms (IA) are abnormal dilatations of a cerebral artery caused by weakening of the vessel wall that occurs due to genetic and acquired factors associated with the mechanical stress of blood flow. According to the World Health Organization (WHO), it affects 10–15 people per 100,000 inhabitants, and its importance relies on the fact that they are considered the most relevant cause of spontaneous subarachnoid hemorrhage. Treatment of ruptured and unruptured aneurysms can be performed by surgical technique (clipping and/or wrapping) or endovascular technique (embolization or flow diverting implantation). The flow diverter implant is based on the blood flow remodeling, which is prevented from entering the aneurysm, resulting in thrombosis of the A1. Postoperative complications include perianeurysmal edema, delayed hemorrhage, and delayed aneurysm rupture, and delayed parent vessel occlusion. However, in this case report, a less common complication, the hyperperfusion syndrome, is presented.

Cerebral hyperperfusion syndrome is a rare condition seen in 0-3% of carotid endarterectomy procedures. It is even rarer when related to a flow diverter implantation procedure. It is caused when a long-term area with hypoperfusion loses its capacity of vascular resistance autoregulation. When reperfusion takes place, this area suffers from uncontrolled augmentation of blood flow. In this case report, a rare condition of hyperperfusion syndrome after a flow diverter implantation procedure for a C4 saccular aneurysm of the internal carotid is presented.

CASE PRESENTATION

A case of a 60-year-old male patient, with a previous history of gastric cancer, referred to a tertiary care service for treatment of an unruptured saccular aneurysm on the cavernous segment of the left internal carotid artery, measuring 2.6 x 4.5 mm (neck x height) is presented (Figure 1). The patient was asymptomatic and the aneurysm was incidentally found on a control brain magnetic resonance imaging (MRI) during follow up of an expansive pituitary lesion. After the initial evaluation, endovascular treatment with flow diverting stent placement was indicated, and the ‘Flow Re-Direction Endoluminal Device’ (FRED®) system was chosen.

The patient was submitted to the procedure under general anesthesia. Right femoral puncture was performed and the passage, positioning and opening of the flow diverter at the site of the saccular aneurysm was uneventful. Control angiography showed delayed contrast retention (Figure 2).

In the immediate postoperative period, the patient developed right hemiparesis and global aphasia. Control computed tomography (CT) showed no bleeding, and digital subtraction angiography (DSA) revealed patency of the arteries. MRI showed no acute alterations that justified clinical manifestations. Patient remained hemodynamically stable under intensive care and was medicated with tirofiban (Figure 3). During hospitalization in the ICU, he developed phenytoin-resistant generalized tonic-clonic seizures, requiring administration of phenobarbital. A new MRI showed cortical left temporo-frontal diffusion restriction, suggestive of hypervascularization in the left hemisphere.

Figure 1. DSA 3D reconstruction: A. saccular aneurysm emerging from cavernous portion of the left internal carotid artery; B. aneurysm neck, height and width dimensions.
Case Report


Four days after the procedure the patient spontaneously evolved with improvement of the aphasia and paresis, being discharged after 10 days without deficits.

DISCUSSION

The hyperperfusion syndrome is a rare condition, mostly seen on endarterectomy procedures or aneurysmal clipping. However, the development of this syndrome after a flow diverter implantation is rarely described. It is logical to infer that the syndrome may also occur with flow diversion and be susceptible to the same predisposing factors as the aneurysm clipping, since the flow-diverting stents reduce blood flow into the aneurysmal sac by directing flow to the distal vessel\(^5,6\). Considering the endothelial lesion involved with the aneurysm, it provokes a failure on myogenic autoregulation function of cerebral vessels, resulting, as a consequence, in hyperperfusion syndrome\(^7\).

Although the pathogenetic mechanisms are not fully understood, it is believed to be caused by brain autoregulation. Long-term hypoperfusion results in chronic compensatory distal blood vessel dilatation, which implicates in loss of the vascular resistance regulation to circulatory changes. When blood flow is recovered, as in procedures such the aforementioned flow diverter implantation, the impaired microvascular autoregulation leads to incapacity of reaction to the recanalization of blood flow and the installation of hyperperfusion syndrome, even if the increase leads to only moderate hyperperfusion\(^6,8\).

The preoperative risk factors most often associated with the development of the syndrome include hypertension, diabetes, age older than 75 years, recent carotid surgery within the past 3 months, high-grade ipsilateral or contralateral stenosis and cerebrovascular reactivity, female sex, vascular malformations.

Figure 2. DSA, lateral view. A. preoperative image; B. flow diverter release moment; C. flow diverter implanted and blood flow restored.

Figure 3. A-D. Foci of acute/subacute ischemia compromising left frontal, parietal and occipital cortex. MRI also showed hypersignal on subcortical FLAIR involving the left frontal lobe and insular cortex. Also, discrete signs of supratentorial ischemic microangiopathy are shown.
and cerebrovascular reactivity. The patient in the present case showed only the recent procedure.

As the patient only had one of the six most prevalent risk factors for the syndrome, the most probable hypothesis is that after the implantation of the flow diverter stent the normal blood flow was reestablished too quickly, which ended up generating a vasogenic edema due to peak of pressure. This edema may be the cause of the motor symptoms and generalized tonic-clonic seizures. According to Narata et al., temporary brain edema is an underrated complication that could be associated to particular procedures such as flow diverter implantation due to stent positioning and artery diameter.

The patient presented with right hemiparesis and expressive aphasia immediately after the postoperative period, evolving to generalized tonic-clonic seizures. The speed of manifestation of the patient's symptoms in the case is in accordance with Garcia-Bargo et al., who addressed that the hyperperfusion syndrome usually occurs in the immediate postoperative period. Besides these symptoms, studies showed that patients may present headaches, visual disturbances, confusional episodes, macular edema, vomit, dysarthria, facial and ocular pain.

CONCLUSION

Hyperperfusion syndrome is a rare, self-limited syndrome with nonspecific clinical manifestations that arise in the postoperative period, mainly after carotid endarterectomy. The description of this syndrome and its relationship with the flow diverter stent are still scarce in the literature, as is the pathophysiology of the disease, which demonstrates the need for further study and deepening on the subject.

REFERENCES


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