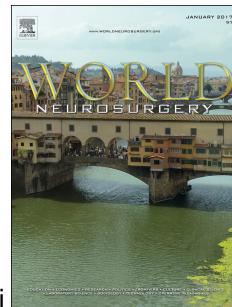


# Accepted Manuscript

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Hyperperfusion syndrome after trapping with high-flow bypass for a giant paraclinoid internal carotid artery aneurysm

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Key words: giant internal carotid artery aneurysm, high-flow bypass, hyperperfusion syndrome

Abbreviations list: ASL-MRI=Arterial spin-labeling magnetic resonance perfusion image; CBF=Cerebral blood flow; CT=Computed tomography; DSA=Digital subtraction angiography; ECD=Ethyl cysteinate dimer; FLAIR=Fluid-attenuated inversion recovery; ICA= Internal carotid artery; MCA=Middle cerebral artery; MRA=Magnetic resonance angiography; MRI=Magnetic resonance imaging; POD= Postoperative day; RA=Radial artery; SPECT=Single photon emission computed tomography.

Informed consent: The patient has consented to the submission of this case report to the journal.

Declaration of interest: none

## Abstract

**Background:** Hyperperfusion syndrome associated with aneurysm surgery is rare. The occurrence of the syndrome after trapping with high-flow bypass has not been described previously. Herein, we present a case of the syndrome occurring after trapping with high-flow bypass of an unruptured giant paraclinoid internal carotid artery (ICA) aneurysm.

**Case Description:** The patient was a 68-year-old woman with progressive loss of vision in her left eye. After a diagnosis of a left giant ICA aneurysm, she underwent successful trapping with high-flow bypass. No new neurological deficits were observed after surgery. Computed tomography (CT) on the same day and magnetic resonance imaging (MRI) on the next day revealed no hemorrhage or infarction. The patient had a headache and transit motor aphasia on postoperative day (POD) 8. Arterial spin-labeling magnetic resonance perfusion image on the same day and single photon emission CT scan on POD 10 demonstrated hyperperfusion in the left cerebral cortex. The symptoms gradually improved over a week and she had no new neurological deficits when discharged from hospital.

**Conclusions:** This report suggests that, although rare, hyperperfusion syndrome after trapping with high-flow bypass should be considered in giant aneurysmal patients if they present with

headache and neurological deficits in a delayed period.

## Introduction

Hyperperfusion syndrome is known to occur after carotid endarterectomy, carotid artery stenting and extracranial-intracranial bypass surgery. The syndrome has also been reported following an aneurysmal surgery for a giant aneurysm after neck clipping and for a large aneurysm after flow diversion.<sup>1-5</sup> However, hyperperfusion syndrome has never been reported after trapping with high-flow bypass. We present a case of hyperperfusion syndrome after trapping with high-flow bypass of an unruptured giant paraclinoid internal carotid artery (ICA) aneurysm.

## Case report

History: The patient, a 68-year-old right-handed woman presented symptoms of progressive loss of vision in her left eye and temporal visual field defect in her right eye. The patient has a history of hypertension and a family history of subarachnoid hemorrhage.

Examination: Magnetic resonance imaging (MRI) revealed a non-thrombotic left paraclinoid ICA giant aneurysm and local compression of the optic chiasma (Fig. 1A). A three-dimensional time of flight magnetic resonance angiography (MRA) study also revealed

an attenuation in ipsilateral anterior circulation as well as the aneurysm (Fig. 1B). Arterial spin-labeling magnetic resonance perfusion image (ASL-MRI) revealed a decrease in cerebral blood flow (CBF) in the left hemisphere (Fig. 1C). Intra-arterial digital subtraction angiography (DSA) demonstrated a left giant paraclinoid ICA aneurysm with poor filling of the left hemisphere branches. DSA revealed that the maximum diameter was 28.8mm (Fig. 2A and B). A balloon occlusion test was performed for the left ICA. The left anterior cerebral artery and middle cerebral artery (MCA) were filling through the anterior communicating artery but not the posterior communicating artery. The aneurysm was visualized through the ophthalmic artery with external carotid angiography. No neurological deficits were observed during the 20-minute occlusion, but venous filling delay had appeared.

**Surgical procedure:** A frontotemporal craniotomy was performed followed by a wide dissection of the Sylvian fissure, and anastomosis of the parietal branch of the superficial temporal artery (STA) to the M4 portion distal to the recipient M2 for insurance during temporary clamping. After the radial artery (RA) graft was passed from the cranium to the neck through the trocar, the RA graft was anastomosed to the M2 main trunk, and then RA-external carotid artery anastomosis was performed. The parent vessel was proximally

occluded after confirming adequate flow in the graft using DSA. The aneurysm was successfully trapped with cervical ICA ligation, aneurysmal distal ICA clipping and ophthalmic artery clipping.

Post-operative course: No neurological deficits were observed post-surgery. CT on the same day and MRI on the next day revealed no hemorrhage or infarction in diffusion-weighted image (DWI) and bypass patency. The patient complained of unilateral headache on the side of the craniotomy on POD 1. The headache was treated with non-steroidal anti-inflammatory drugs. No abrupt elevation of blood pressure was observed post-operatively and the patient's blood pressure remained normotensive. Intra-arterial DSA after surgery demonstrated good patency of the bypasses and disappearance of flow to the aneurysm (Fig. 3A and B). The patient had 60 minutes transit motor aphasia on POD 8, and the MRI obtained on the same day demonstrated no infarction in DWI. However, ASL-MRI revealed an increase in CBF in the left hemisphere and fluid-attenuated inversion recovery (FLAIR) demonstrated a hyperintense area in the left cortex (Fig. 4A and B). The patient was administered an anti-hypertensive drug and the patient's blood pressure was monitored closely and remained normotensive. Electroencephalography obtained on POD 9 did not show spike waves. CBF

using Tc-99m L, L-ethyl cysteinate dimer (ECD) single photon emission computed tomography (SPECT) scan on POD 11 revealed an approximately 15-30% increase in the regional CBF of the left frontotemporoparietal lobe compared to that of the contralateral hemisphere (Fig. 5A and B). The patient's headache and transit motor aphasia improved on POD 16, and CBF using ECD SPECT scan on POD 18 improved, showing an approximately 6-13% increase in regional CBF of the left hemisphere compared to the contralateral hemisphere. The patient had no symptoms when discharged from hospital.

## Discussion

Giant aneurysm has a dismal natural history and often results in hemorrhage, cerebral compression, and thromboembolism. Giant aneurysms of the intracranial ICA frequently involve the parent artery and critical neighboring branches, and direct surgical clip reconstruction is difficult in many cases. Trapping with high-flow bypass can completely exclude giant ICA aneurysms from the flow stream promptly inducing thrombosis inside the aneurysm. Ischemia resulting from the bypass graft occlusion has been reported as a complication of trapping with high-flow bypass; however, hyperperfusion syndrome has never been reported to cause focal neurological signs after trapping with high-flow bypass for

aneurysmal obliteration.<sup>6</sup>

Patients with hyperperfusion syndrome typically present with symptoms, such as, severe headache, vomiting, confusion, seizure, and other neurological signs of increased intracranial pressure. The syndrome in aneurysmal cases has been reported to develop as severe headache, aphasia, and hemiparesis after undergoing clipping and endovascular flow diversion of an unruptured cerebral aneurysm. The syndrome developed between the time immediately after surgery and the ninth day of the surgery.<sup>1-5</sup> In our patient, hyperperfusion syndrome developed as aphasia on POD 8.

We examined whether hypoperfusion of the preoperative cerebral hemisphere, indicated by ASL-MRI, is a real phenomenon or an artifact of delayed transit time caused by stagnation of turbulent flow within the giant aneurysm. In this giant cerebral aneurysm case, MRA showed attenuation in ipsilateral anterior circulation. Cerebral blood flow measured before surgery was reported as hypoperfusion.<sup>1</sup> We agree with the report and think that it was actually hypoperfusion and not merely an artifact.

Hyperperfusion syndrome is thought to occur when perfusion is rapidly increased in a chronically hypoperfused area. MR perfusion study showed impaired hemodynamics in the

vascular territory supplied by MCA of the aneurysm side in 4 out of 5 patients harboring intracranial ICA giant aneurysms.<sup>7</sup> In our patient, ischemic symptoms were not observed before surgery; however, ASL-MRI revealed a decrease in CBF on the side of the aneurysm. For this reason, our case with decreased blood flow showed improved flow after high-flow bypass which led to the syndrome.

### Conclusion

When a giant ICA aneurysm, wherein blood flow is reduced on the side of the aneurysm, is trapped with high-flow bypass, the increase in blood flow through the bypass may lead to hyperperfusion syndrome. Post-operative SPECT and ASL-MRI may be useful in identifying potential cases of hyperperfusion syndrome.

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## Figure Legends

Figure 1. Pre-operative images. A: MRI coronal view showing the nonthrombotic giant aneurysm and local compression of the optic chiasma. B: MRA showing the left ICA aneurysm and attenuation in ipsilateral anterior circulation. C: ASL-MRI obtained before the operation revealed a decreased signal in the left hemisphere. The aneurysm is depicted as a hyperintense signal.

Figure 2. Pre-operative images. A and B: Frontal and lateral DSA views of the left ICA revealing a giant paraclinoid aneurysm with poor filling of the normal hemisphere branch.

Figure 3. Post-operative images. A and B: DSA performed after surgery demonstrating good patency of the bypasses and disappearance of flow to the aneurysm.

Figure 4. Post-operative image. A: FLAIR MRA obtained on post-operative day 8 demonstrating hyperintense areas in the left frontoparietal area. B: ASL-MRI obtained on post-operative day 8 revealed an increased signal in the left hemisphere.

Figure 5. A and B: Tc-99m ECD SPECT scan obtained on post-operative day 10 demonstrating increased regional blood flow in the left hemisphere.

Fig. 1

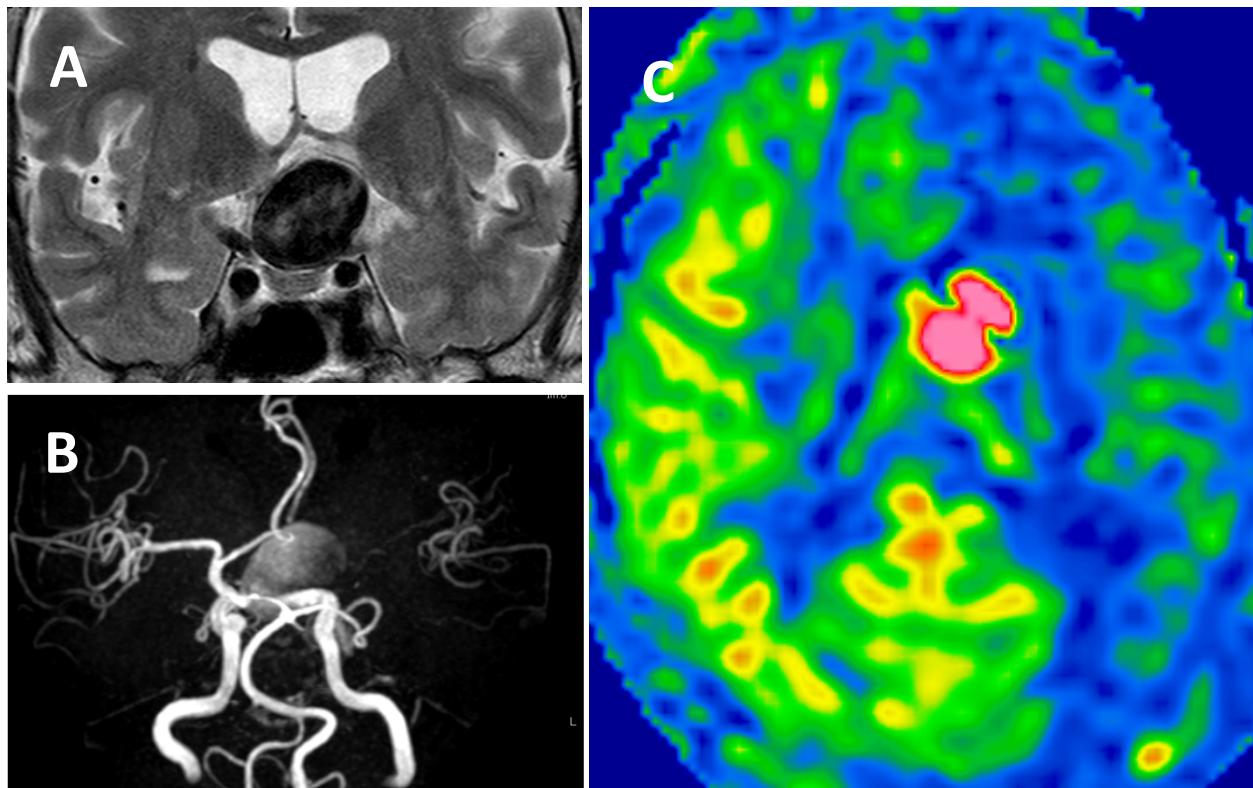


Fig. 2

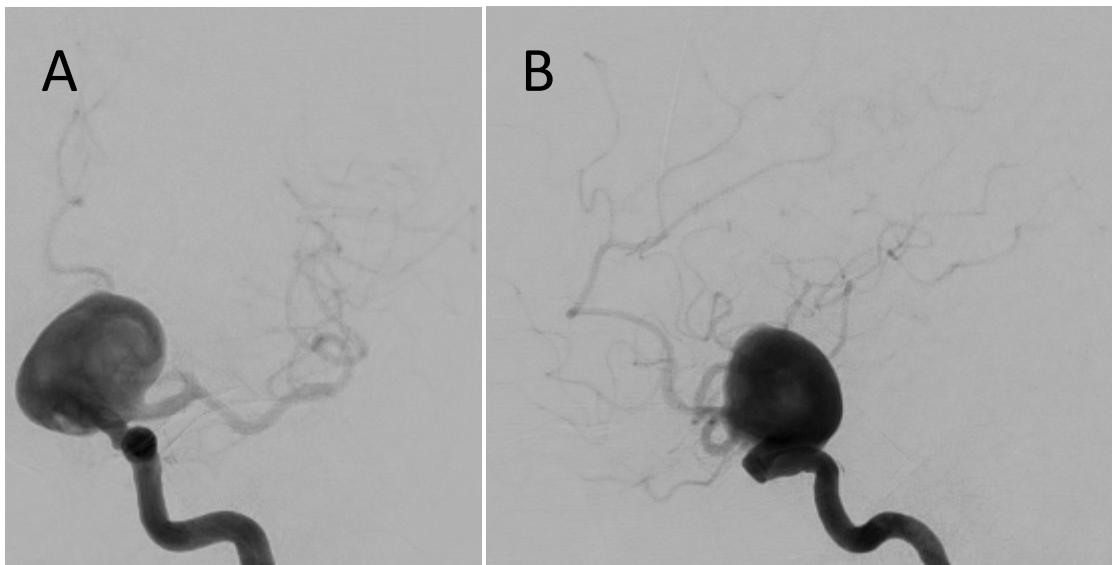


Fig. 3

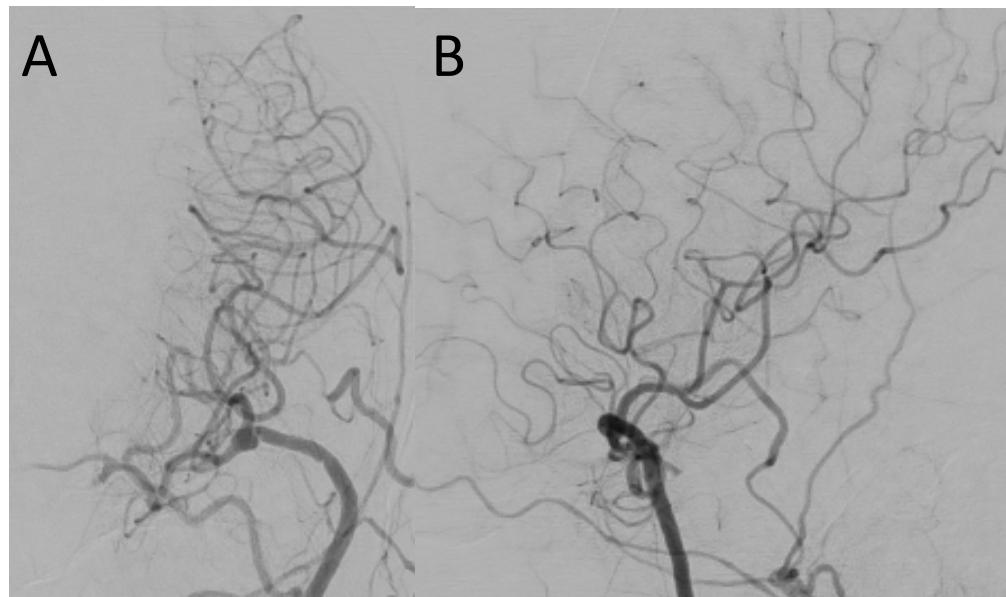


Fig. 4

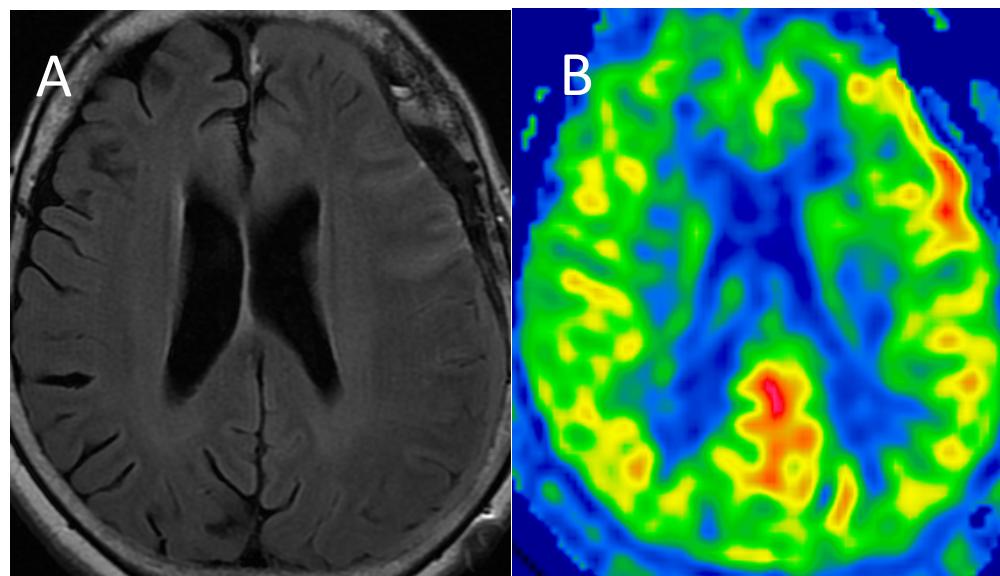
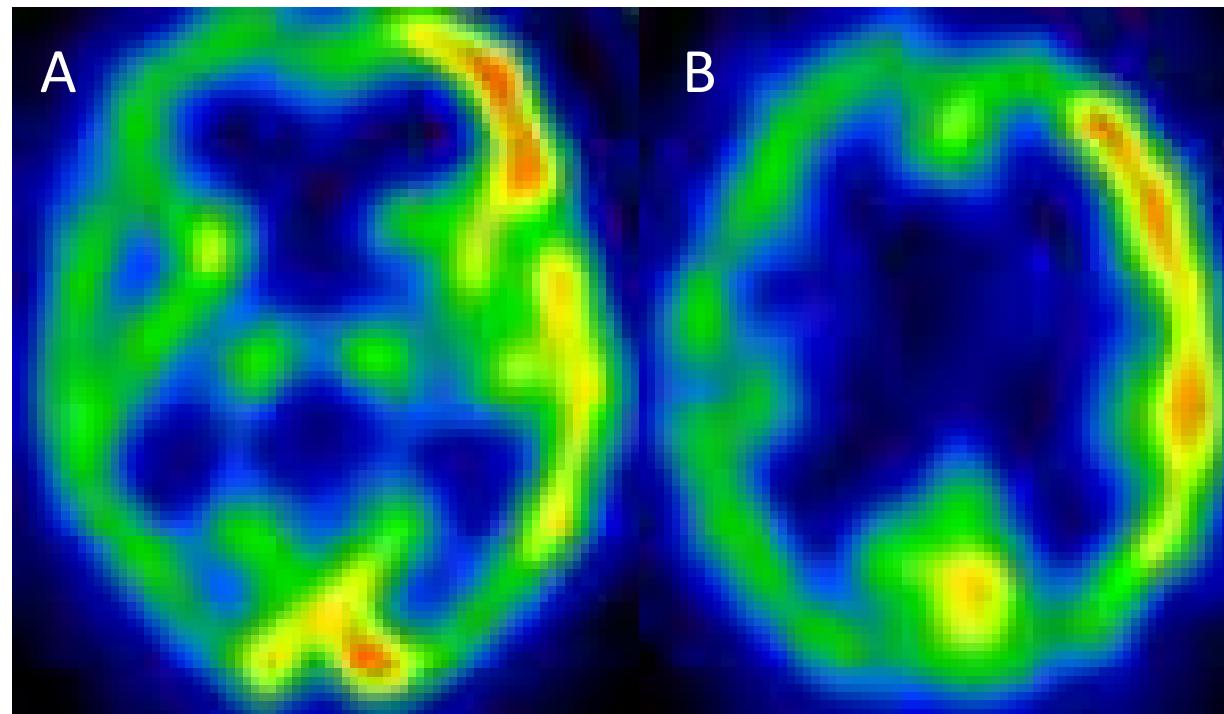


Fig. 5



Highlight

- Giant ICA aneurysm may have decreased cerebral blood flow on the side of the aneurysm
- Hyperperfusion syndrome after high-flow bypass surgery for cerebral aneurysm
- Post-operative SPECT and ASL-MRI may be useful in identifying potential cases of hyperperfusion syndrome