Transient Crossed Cerebellar Diaschisis Due to Cerebral Hyperperfusion Following Surgical Revascularization for Moyamoya Disease

—Case Report—

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Abstract

Crossed cerebellar diaschisis (CCD) often occurs after ischemic or hemorrhagic stroke that damages the cortico-ponto-cerebellar pathway. However, CCD due to cerebral hyperperfusion following cerebrovascular reconstruction is rare. A 61-year-old woman presented with transient CCD due to cerebral hyperperfusion following bypass surgery for adult moyamoya disease. She developed transient weakness of the right extremities and was diagnosed with moyamoya disease. First, she underwent superficial temporal artery to middle cerebral artery (STA-MCA) anastomosis with indirect synangiosis on the left. Postoperative course was uneventful. Subsequently, she underwent STA-MCA anastomosis with indirect synangiosis on the right. She complained of mild headache on the right, and single photon emission computed tomography (SPECT) performed on the 7th postoperative day demonstrated hyperperfusion in the right frontal and temporal lobes associated with hypoperfusion in the left cerebellum. Magnetic resonance (MR) imaging demonstrated no new lesions and MR angiography showed patent STA-MCA bypass. Subsequent SPECT showed disappearance of both hyperperfusion and CCD. This case strongly suggests that cerebral hyperperfusion after bypass surgery for moyamoya disease may cause transient CCD. Although the clinical significance is still obscure, this phenomenon indicates the cortico-ponto-cerebellar pathway is interrupted due to hyperperfusion, suggesting the development of hyperperfusion syndrome. Careful observation of cerebral hemodynamics after bypass surgery is warranted to avoid hyperperfusion-related complications.

Key words: moyamoya diseases, crossed cerebellar diaschisis, hyperperfusion, bypass surgery, cerebral blood flow

Introduction

Crossed cerebellar diaschisis (CCD) is defined as the reduction of metabolism and blood flow in the cerebellar hemisphere contralateral to supratentorial lesions. This unique phenomenon was first characterized using positron emission tomography (PET).2) CCD is frequently observed in patients with cerebral infarction or intracerebral hemorrhage in the middle cerebral artery (MCA) territory presenting hemiparesis. This remote functional depression is believed to occur due to the interruption of the cortico-ponto-cerebellar pathway.2,16,22) However, CCD can occur in patients without motor deficits such as hemiparesis, and is sometimes absent in hemiparetic patients.16,17,22) Therefore, the specific mechanisms underlying CCD are still obscure. Cerebral blood flow (CBF) is decreased in the contralateral cerebral hemisphere in most patients with CCD.12)

Cerebral hyperperfusion is known to occur after revascularization surgery, including carotid endarterectomy (CEA) and superficial temporal artery to MCA (STA-MCA) anastomosis.15,20,21,23,24,26) In particular, cerebral hyperperfusion syndrome occurs in about 30% of adult patients with moyamoya disease after STA-MCA anastomosis.5) However, there are few reports of CCD due to cerebral hyperperfusion after bypass surgery. Only one case developed transient CCD after CEA.19)

We report a case of transient CCD secondary to cerebral hyperperfusion following bypass surgery for adult moyamoya disease.

Case Report

A 61-year-old female developed transient weakness of the...
right extremities and was admitted to our hospital. She had a past history of hypertension, diabetes mellitus, and hyperlipidemia. Magnetic resonance (MR) imaging revealed no fresh cerebral infarctions, but MR angiography and digital subtraction angiography demonstrated bilateral occlusive changes in the terminal portions of the internal carotid arteries associated with an abnormal vascular network at the base of the brain (Fig. 1). Therefore, the diagnosis was moyamoya disease. N-isopropyl-p-[123I]iodoamphetamine ([123I-IMP]) single photon emission tomography (SPECT) showed marked reduction of CBF and cerebrovascular reactivity (CVR) to acetazolamide in the bilateral frontal and temporal lobes (Fig. 2A, B). First, she underwent STA-MCA anastomosis with indirect syn-angiosis, encephalo-duro-myo-arterio-pericranio-synangiosis (EDMAPS) on the left. Postoperative course was uneventful, and she developed no further ischemic attacks.

Preoperative [123I-IMP] SPECT demonstrated distinct improvements of CBF and CVR in the left cerebral hemisphere, but persistent decreases in CBF and CVR on the right (Fig. 2C, D). Therefore, she underwent STA-MCA anastomosis and EDMAPS on the right 17 months after the first surgery. She developed no neurological deficits after surgery. Systolic blood pressure was controlled between 100 and 140 mmHg to prevent postoperative hyperperfusion syndrome, using continuous infusion of nicardipine. [123I-IMP] SPECT on the 2nd postoperative day showed mild improvement of CBF in the right hemisphere.

Fig. 1  A: Fluid-attenuated inversion recovery magnetic resonance image revealing an old small infarction in the left corona radiata, but no acute cerebral infarction. B, C: Right (B) and left internal carotid angiograms (C), Towne's view, demonstrating severe stenosis in the terminal portions of the bilateral internal carotid arteries associated with an abnormal vascular network at the base of the brain, suggesting moyamoya disease.

Fig. 2  Serial N-isopropyl-p-[123I]iodoamphetamine single photon emission tomography (SPECT) scans showing cerebral blood flow (CBF) at rest (A) and cerebrovascular reactivity (CVR) to acetazolamide (B) were reduced in the bilateral frontal and temporal lobes before surgery, and both CBF (C) and CVR (D) in the left hemisphere significantly improved after superficial temporal artery to middle cerebral artery (STA-MCA) anastomosis and indirect bypass on the left. Note the persistent decrease in CBF and CVR in the right hemisphere. Two days after STA-MCA anastomosis and indirect bypass on the right, CBF in right hemisphere improved (E) with no laterality in the cerebellar hemispheres (F). Note postoperative hyperperfusion in the right frontal and temporal lobes (arrows in G) associated with blood flow reduction in the left cerebellum (arrowhead in H) on the 7th postoperative day. Postoperative hyperperfusion (I) and crossed cerebellar diaschisis (J) completely disappeared on the 14th postoperative day.

Fig. 3  A: Postoperative fluid-attenuated inversion recovery magnetic resonance (MR) image demonstrating no new lesion. B: MR angiogram showing patent superficial temporal artery to middle cerebral artery anastomosis on both sides (arrows).
(Fig. 2E) and no laterality in the cerebellar hemispheres (Fig. 2F). Subsequently, she complained of mild headache on the right. $^{123}$I-IMP SPECT on the 7th postoperative day demonstrated postoperative hyperfusion in the right frontal and temporal lobes (Fig. 2G) and marked reduction of blood flow in the left cerebellar hemisphere (Fig. 2H).

Thus, careful neurological observations and control of blood pressure were continued. Her headache resolved within a few days. $^{123}$I-IMP SPECT on the 14th postoperative day demonstrated disappearance of the hyperperfusion in the right hemisphere (Fig. 2I) and CCD (Fig. 2J). Postoperative MR imaging demonstrated no new lesion and MR angiography showed the patent STA-MCA bypass (Fig. 3). She was discharged 3 weeks after surgery without neurological deficits. No cerebrovascular events occurred during the follow-up period.

**Discussion**

This is a very rare case of transient CCD associated with cerebral hyperperfusion after surgical revascularization for adult moyamoya disease. Previous studies have revealed that preoperative persistent dilation of resistance vessels is the most likely cause of postoperative hyperperfusion after surgical revascularization such as CEA and STA-MCA anastomosis. Hyperperfusion syndrome includes headache, seizure, focal neurological deficits, and intracerebral hemorrhage, and is sometimes lethal. Nowadays, such vascular conditions can be identified as a decrease in cerebral perfusion reserve, using SPECT and PET, and may be a significant predictor for postoperative hyperperfusion. Blood flow study immediately after surgery would also be beneficial to detect postoperative hyperperfusion and prevent hyperperfusion-related neurological complications caused by sedation and blood pressure control. Recent evidence suggests that surgical revascularization for moyamoya disease may often cause symptomatic cerebral hyperperfusion, with a reported incidence of about 20% to 30%, higher than that after STA-MCA anastomosis for atherosclerotic disease. Poor vascular networks in moyamoya disease may explain why patients with moyamoya disease have higher risk for postoperative hyperperfusion.

However, cerebral hyperperfusion following surgical revascularization may rarely cause transient CCD. Actually, we treated 26 patients (40 sides) with surgical revascularization for adult-onset moyamoya disease at our hospital between April 2006 and July 2011, including this patient. During the period, all patients underwent serial SPECT study conducted on postoperative days 0, 2, and 7. In those adult-onset moyamoya patients, symptomatic hyperperfusion was observed in 12 (30%) of 40 hemispheres and asymptomatic hyperperfusion in 14 (35.0%) hemispheres. However, the present patient was the only case with CCD due to cerebral hyperperfusion. Previously, only one case had developed transient CCD due to hyperperfusion after CEA. This patient demonstrated cerebral hyperperfusion and CCD 3 days after CEA, and developed neurological deficits on the 4th postoperative day despite intensive control of blood pressure. Therefore, the preceding CED may suggest the development of occult hyperperfusion syndrome as the result of reduction of cerebral metabolism due to cerebral hyperperfusion.

Generally, interruption of the cortico-ponto-cerebellar pathway causes CCD. Corticopontine fibers are present in nearly the entire cerebral cortex and are in close contact with, but are more spread out than the cortico-spinal fibers. In addition, corticopontine fibers from the association cortex are twenty times more abundant than those originating from the primary motor cortex. Furthermore, a PET study has revealed that the frontal cortex, particularly the premotor and prefrontal areas, exerts a strong modulating influence on the metabolism in the contralateral cerebellum in humans. Therefore, postoperative hyperperfusion may functionally suppress the relatively wide cerebral cortex and reduce metabolic activity in the contralateral cerebellar hemisphere as a remote effect (diaschisis). The present case had only mild headache due to postoperative hyperperfusion, but serial radiological findings strongly suggested that excess blood flow in the right cerebral hemisphere may have caused metabolic depression in the left cerebellum through the cortico-ponto-cerebellar pathway, leading to transient CCD. Although this phenomenon is rarely reported, further studies are needed to clarify whether hyperperfusion-related CCD can be a predictor for the development of hyperperfusion syndrome.

The findings in the present case strongly suggest that cerebral hyperperfusion after bypass surgery for moyamoya disease may cause transient CCD. Although the clinical significance is still obscure, this phenomenon indicates the cortico-ponto-cerebellar pathway is interrupted due to postoperative hyperperfusion, suggesting the development of hyperperfusion syndrome. Postoperative critical care and careful observations of cerebral hemodynamics are recommended to prevent hyperperfusion syndrome.

**References**

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