

## Local Vasogenic Edema without Cerebral Hyperperfusion after Direct Revascularization Surgery for Moyamoya Disease

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Superficial temporal artery–middle cerebral artery anastomosis is generally used as the standard surgical treatment for moyamoya disease to prevent cerebral ischemic attacks. Although the main potential complications associated with this treatment are cerebral hyperperfusion and ischemia, the adverse impacts of revascularization surgery remain unclear. Of the 142 consecutive surgeries for moyamoya disease at our hospital from 2008, we herein presented 2 cases of adult-onset moyamoya disease that manifested local vasogenic edema at the site of anastomosis without cerebral hyperperfusion; 1 in a 31-year-old woman presented with transient ischemic attack and the other in a 22-year-old man manifested as minor completed stroke. Both patients underwent superficial temporal artery–middle cerebral artery anastomosis, resulting in the formation of a reversible high-signal-intensity lesion at the site of anastomosis on T2-weighted images along with an increase in apparent diffusion coefficient values, whereas diffusion-weighted images showed no changes. Neither hyperperfusion nor hypoperfusion, as assessed by single-photon emission computed tomography with N-isopropyl[123I]-p-iodoamphetamine, was observed postoperatively. In light of the increased risk of the further progression of vasogenic edema to intracerebral hemorrhage, these patients were treated with prophylactic blood pressure lowering and the intravenous infusion of a free radical scavenger. They did not have any further cerebrovascular events during the follow-up period. Regional vasogenic edema without cerebral hyperperfusion, possibly due to cerebral ischemia/reperfusion injury, may be another novel entity that needs to be considered as a potential complication after extracranial–intracranial bypass for moyamoya disease. Strict postoperative management should be used to avoid hemorrhagic transformation. **Key Words:** Moyamoya disease—vasogenic edema—reperfusion—cerebral hyperperfusion—extracranial–intracranial bypass.

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Received November 17, 2014; revision received January 28, 2015; accepted March 18, 2015.

There is no grant support.

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1052-3057/\$ - see front matter

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<http://dx.doi.org/10.1016/j.jstrokecerebrovasdis.2015.03.050>

## Introduction

Moyamoya disease is a chronic, occlusive cerebrovascular disease of unknown etiology that is characterized by bilateral steno-occlusive changes at the terminal portion of the internal carotid artery and an abnormal vascular network at the base of the brain.<sup>1</sup> Extracranial-intracranial bypass such as superficial temporal artery-middle cerebral artery (STA-MCA) anastomosis is generally used as the standard surgical treatment for moyamoya disease to prevent cerebral ischemic attacks.<sup>2,3</sup> Although the long-term outcome of STA-MCA anastomosis is favorable, potential complications in the early postoperative period have been reported. Focal cerebral hyperperfusion causes transient neurologic deterioration or delayed intracerebral hemorrhage during the acute stage after revascularization for moyamoya disease.<sup>4,5</sup> Another potential complication that has been reported, especially in younger patients, is the watershed shift, which induces ischemic lesions in the remote territory of MCA because of a change in the flow pattern after STA-MCA anastomosis.<sup>6</sup> These opposite phenomena associated with the dynamic changes in cerebral hemodynamics make the postoperative management of moyamoya disease complicated. Furthermore, the adverse impacts of revascularization surgery in moyamoya disease remain unclear.

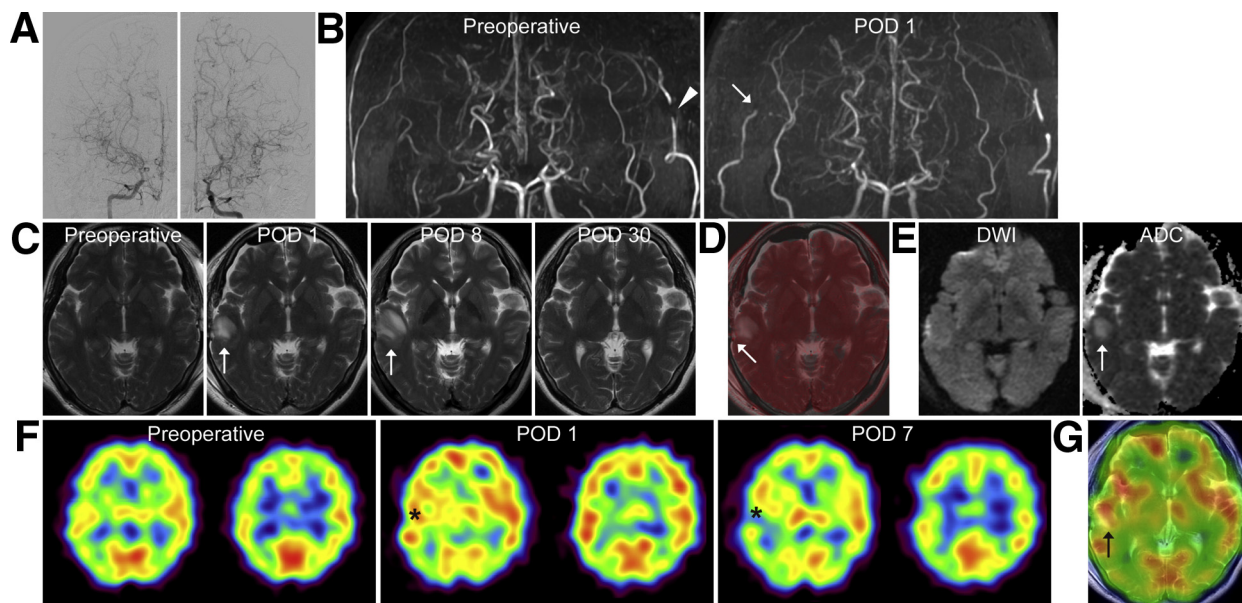
Of the 142 consecutive surgeries performed for moyamoya disease by the same surgeon under a standardized protocol (postoperative prophylactic intensive blood pressure control [ $<130$  mm Hg systolic blood pressure]) at our hospital from January 2008,<sup>7</sup> we herein presented 2 cases of adult-onset moyamoya disease, which manifested local vasogenic edema at the site of anastomosis without cerebral hyperperfusion after STA-MCA anastomosis. This apparently unique entity may be another potential complication of STA-MCA anastomosis for moyamoya disease.

## Case Reports

### Case 1

#### History and Examination

A 31-year-old woman presented with crescendo transient motor weakness in her right upper extremity. Cerebral angiography revealed occlusive changes at the terminal portion of the bilateral internal carotid arteries associated with abundant moyamoya vessels on both sides, leading to a diagnosis of moyamoya disease (Fig 1, A). Because initial single-photon emission computed tomography with N-isopropyl[123I]-p-iodoamphetamine ( $^{123}\text{I}$ -IMP-SPECT) revealed an apparent decrease in cerebral blood flow (CBF) on the left



**Figure 1.** Case 1: (A) Preoperative bilateral carotid angiograms revealing occlusive changes at the terminal portion of the bilateral internal carotid arteries along with moyamoya vessels. (B) Magnetic resonance angiography showing the patency of left STA-MCA bypass preoperatively (arrow head) and right STA-MCA bypass 1 day after the second surgery (arrow). (C) Temporal profile of T2-weighted images demonstrating a high-signal-intensity lesion in the right temporal lobe at the site of anastomosis 1 and 8 days after the second surgery (arrows), which completely disappeared 30 days after surgery. (D) A merged image of the T2-weighted magnetic resonance image and the corresponding slice of magnetic resonance angiography (red), 1 day after surgery, showing that the high-signal-intensity lesion existed at the site of anastomosis (arrow). (E) In contrast to no obvious changes being observed on diffusion-weighted images (DWIs), apparent diffusion coefficient (ADC) values increased at the high-signal-intensity lesion 1 day after the second surgery (arrow). (F) Single-photon emission computed tomography with N-isopropyl[123I]-p-iodoamphetamine showing no signs of cerebral hyperperfusion at the site of anastomosis after revascularization surgery (asterisks). (G) A fused image of T2-weighted magnetic resonance image and single-photon emission computed tomography with N-isopropyl[123I]-p-iodoamphetamine 1 day after surgery. Arrow indicates the high-signal-intensity lesion. Abbreviations: POD, postoperative day; STA-MCA, superficial temporal artery middle cerebral artery.

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