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Angiographic characteristics of improper watershed shift after STA-MCA bypass in a patient with moyamoya disease: illustrative case

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BACKGROUND In patients with moyamoya disease (MMD) who receive superficial temporal artery (STA) to middle cerebral artery (MCA) bypass, hypoperfusion remote from the anastomosis site rarely occurs. Watershed shift due to direct bypass has been proposed as the mechanism; however, no report has confirmed this phenomenon using angiography.

OBSERVATIONS A 48-year-old man presented with transient weakness in his left arm. Angiography revealed severe bilateral stenosis of the MCAs and moyamoya vessels. The right anterior cerebral artery (ACA) had short stenosis at A2 but ample blood supply to the cortical area of the right ACA and MCA regions. The patient was diagnosed with MMD and received a single STA-MCA bypass. The next day, he had difficulty communicating, and a cerebral infarction away from the anastomosis site was identified. Perfusion examination revealed hyperperfusion around the direct bypass and hypoperfusion away from the anastomosis site. Angiography revealed bypass patency; however, the original anterograde flow of the right ACA decreased significantly at the stenosed point, indicating an improper watershed shift.

LESSONS STA-MCA bypass for patients with MMD can cause an improper watershed shift decreasing cerebral flow. Donor flow should be prepared based on each angiographic characteristic, and the risk of the improper watershed shift should be considered.

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KEYWORDS moyamoya disease; watershed shift; angiography

Superficial temporal artery (STA) to middle cerebral artery (MCA) bypass combined with indirect revascularization is widely performed in ischemic patients with moyamoya disease (MMD).[1,2] Hyperperfusion is an important cause of postoperative neurological deterioration due to an immediate increase in cerebral blood flow (CBF) after STA-MCA bypass.[3] In contrast, although there have been few reports, hypoperfusion remote from the anastomosis site also leads to postoperative ischemic symptoms.[4] The shift in the watershed region due to direct bypass has been speculated to be the mechanism for this phenomenon;[4,5] however, no study has clearly supported this hypothesis based on angiographic images. Herein, we report the case of a patient with MMD who experienced cerebral infarction after STA-MCA anastomosis. Cerebral perfusion images showed a hypoperfusion area away from the direct bypass, and angiography revealed a shift in the watershed region to an improper boundary.

Illustrative Case

A 48-year-old man presented with transient weakness in the left upper extremity. Magnetic resonance imaging (MRI) revealed multiple acute small infarctions in the right MCA territory on diffusion-weighted imaging (DWI). Magnetic resonance angiography (MRA) revealed severe bilateral occlusion at the origin of the MCAs. N-isopropyl-p-[^{123}I]

ABBREVIATIONS ACA = anterior cerebral artery; CBF = cerebral blood flow; DSA = digital subtraction angiography; DWI = diffusion-weighted imaging; ^{123}I-IMP SPECT = [^{123}I] iodoamphetamine single-photon emission computed tomography; MCA = middle cerebral artery; MMD = moyamoya disease; MRA = magnetic resonance angiography; MRI = magnetic resonance imaging; PCA = posterior cerebral artery; STA = superficial temporal artery.

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iodoamphetamine single-photon emission computed tomography ([123]I-IMP SPECT) under resting conditions showed normal CBF, but those with acetazolamide infusion demonstrated a decrease in CBF in the right MCA region (Fig. 1). Digital subtraction angiography (DSA) revealed severe bilateral stenosis at the origin of the MCA and moyamoya vessels at the brain base. The right anterior cerebral arteries (ACAs) developed and widely supplied blood flow to the cortical area of the right ACA and MCA regions; however, short stenosis with a medium degree was observed at the proximal portion of the A2 segment. In addition, the right posterior cerebral arteries (PCAs) showed stenosis and relatively small perfusion areas (Fig. 2). Based on these radiological findings, the patient was diagnosed with ischemic MMD.

To prevent further ischemic events, the patient received a single STA-MCA anastomosis combined with encephalo-duro-myo-synangiosis in the right MCA territory. Immediately after surgery, the patient was alert and showed no neurological deficits. On the next day at noon, although bypass patency was confirmed by contrast-enhanced computed tomography, he could not maintain rest and had difficulty communicating due to slur and language preservation. On MRI performed 2 days after the operation, acute infarction was identified in the right inferior parietal lobule and white matter of the right frontal lobe. MRA showed the development of STA-MCA bypass; the diameter of the STA was larger than that preoperatively (Fig. 3A–C). As for perfusion examination, [123]I-IMP SPECT showed a significant CBF increase around the anastomosis site of the STA-MCA bypass. Conversely, CBF in the right MCA and ACA regions, remote from the anastomosis site, decreased significantly (Fig. 3D). To determine the cause of this hemodynamic change, we performed angiography. On DSA (Fig. 4), the STA-MCA bypass covered approximately one-half of the right MCA region. However, the blood flow of the right ACA, which widely supplied blood flow to the cortical area of the right ACA and MCA regions before surgery, was remarkably decreased distal to the stenosed point at the proximal portion of A2. The perfusion area supplied by the right PCA remained unchanged. On common carotid artery angiography, the regions corresponding to the right inferior parietal lobule and white matter of the right frontal lobe were avascular.

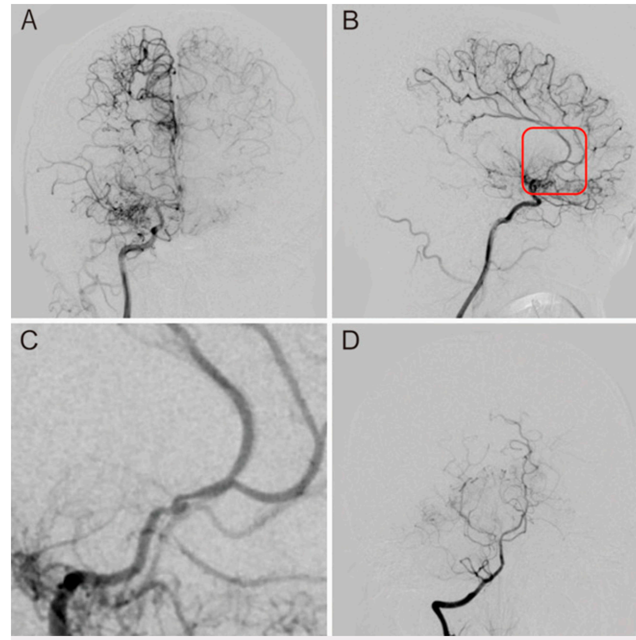

FIG. 2. Preoperative DSA. Right internal carotid artery angiograms, anteroposterior view (**A**) and lateral view (**B**), showed the severe stenosed MCA and moyamoya vessels. In addition, the right ACA was developed and widely covered cortical areas of the right ACA and MCA regions. Enlarged image (**C**) of the area enclosed by the *red square* in panel B. The short stenosis with a medium degree was observed at the proximal portion of the A2 segment. Right vertebral artery angiogram, anteroposterior view (**D**), showed medium stenosis of the right PCA and its relatively small perfusion area.

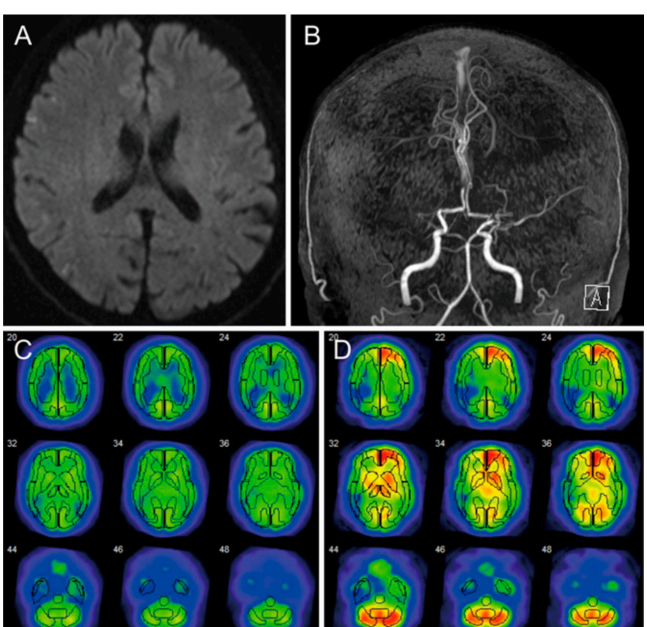

FIG. 1. Preoperative radiological examinations. **A:** DWI identified the acute multiple small infarctions in the right MCA region. **B:** MRA showed bilateral severe stenosis at the origin of MCAs. **C:** SPECT under the rest condition showed the normal CBF. **D:** SPECT with acetazolamide infusion demonstrated the CBF decrease in the right MCA region.

Regarding treatment, because hyperperfusion around the direct bypass and hypoperfusion away from the anastomosis site coexisted, we kept the blood pressure normal and infused a free radical scavenger. One week after surgery, no additional infarction was observed on DWI, and the patient's restlessness and slur gradually improved. Three weeks after surgery, the hyperperfusion around the direct bypass improved, but hypoperfusion in the right frontal lobe remained. Detailed neurological examination revealed executive dysfunction related to the right frontal lobe, and the patient was transferred to a rehabilitation hospital. We will continue to follow up with the expectation of the development of indirect bypass.

Discussion

Observations

We describe a case of MMD in a patient who experienced cerebral infarction after an STA-MCA bypass. Cerebral perfusion examination showed increased CBF around the anastomosis site and decreased CBF remotely from the direct bypass. On angiography, due to the direct bypass, the supply pattern of CBF in the right hemisphere changed dynamically. The STA-MCA bypass covered approximately half of the right MCA region (Fig. 4A and B); however, the original

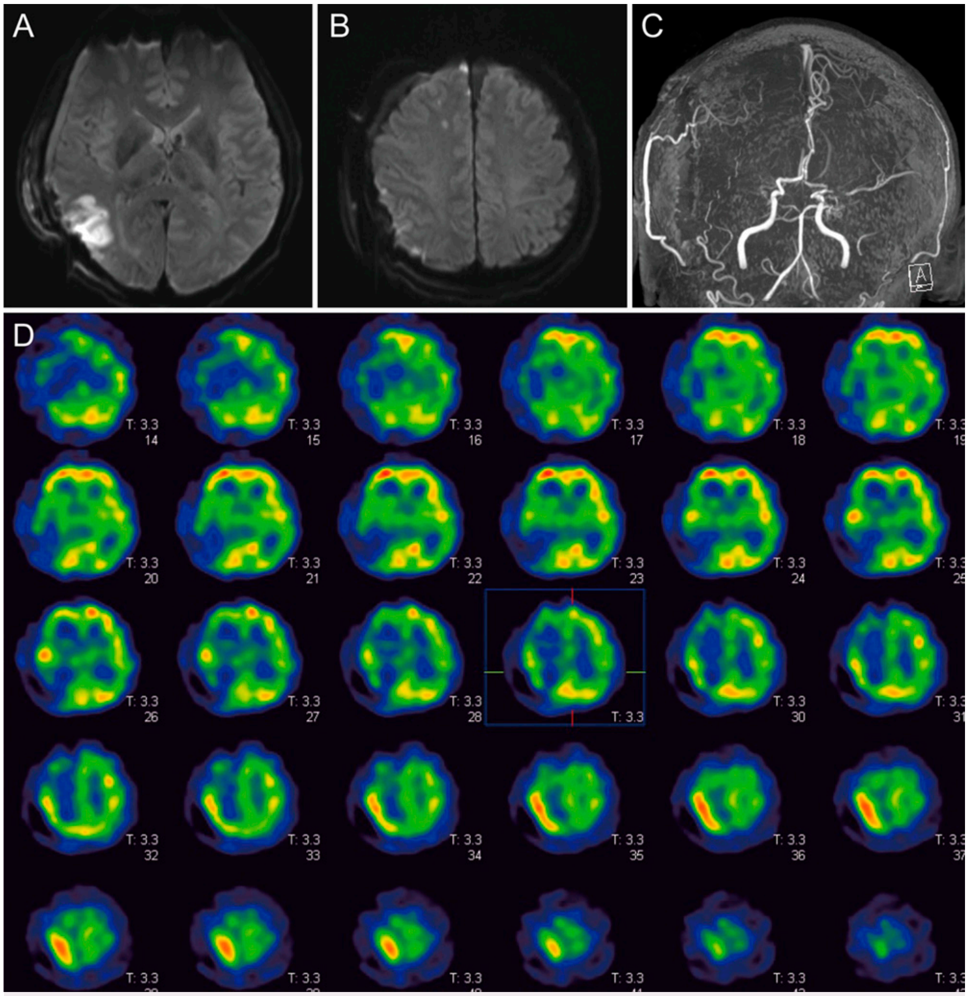

FIG. 3. Postoperative radiological examinations. **A:** DWI showed acute cerebral infarction of the right inferior parietal lobule. **B:** DWI also identified multiple small infarctions in the white matter of the right frontal lobe. **C:** MRA showed the apparent development of STA-MCA bypass. **D:** SPECT under the rest condition revealed the significant CBF increase around the anastomosis site of STA-MCA bypass. However, CBF in the right MCA and ACA regions remote from the anastomosis site was decreased widely.

cerebral flow from the right ACA decreased significantly (Fig. 4C and D). Comparing the preoperative internal carotid artery angiograms (Fig. 2A and B) and postoperative common carotid artery angiograms (Fig. 4G and H), avascular areas that were remote from the direct bypass appeared. These angiographic characteristics indicate that the shift in the watershed area by direct bypass was not adjusted properly to completely cover the preoperative CBF. In patients with MMD after STA-MCA bypass, hypoperfusion away from the anastomosis site of the direct bypass was not common, and a few studies have reported this phenomenon. Hayashi et al.[4,6] proposed a mechanism in which reverse flow by the STA-MCA bypass decreases original anterograde flow and induces relative hypoperfusion in the remote territory of the MCA.[4] This theory is convincing and widely accepted; however, it has remained speculative because of the lack of early postoperative angiography. To the best of our knowledge, this is the first report to clearly represent this phenomenon using angiography.

Regrettably, in this case, cerebral infarction occurred due to an improper shift in the watershed area by direct bypass. Therefore, the cause should be assessed, and treatment strategies should be improved. A noteworthy characteristic of preoperative angiography was that the right ACA widely covered the cortical area of the right ACA and MCA regions but had a short stenosed part in the proximal portion of A2 (Fig. 2A and B). After surgery, the right ACA supplied only a small amount of blood flow distal to the stenosed portion (Fig. 4C–E), which indicated that anterograde flow from the ACA and reverse flow from the STA-MCA bypass conflicted immediately after the stenosed point. However, a single STA-MCA bypass was not sufficient to compensate for the original perfusion area of the right ACA, resulting in cerebral infarction. We speculate that the anterograde flow of the right ACA just after the stenosis was weak and easily defeated by the reverse flow of the direct bypass. Therefore, for revascularization surgery for patients with such a specific vascular structure, considering the occlusion risk of the original

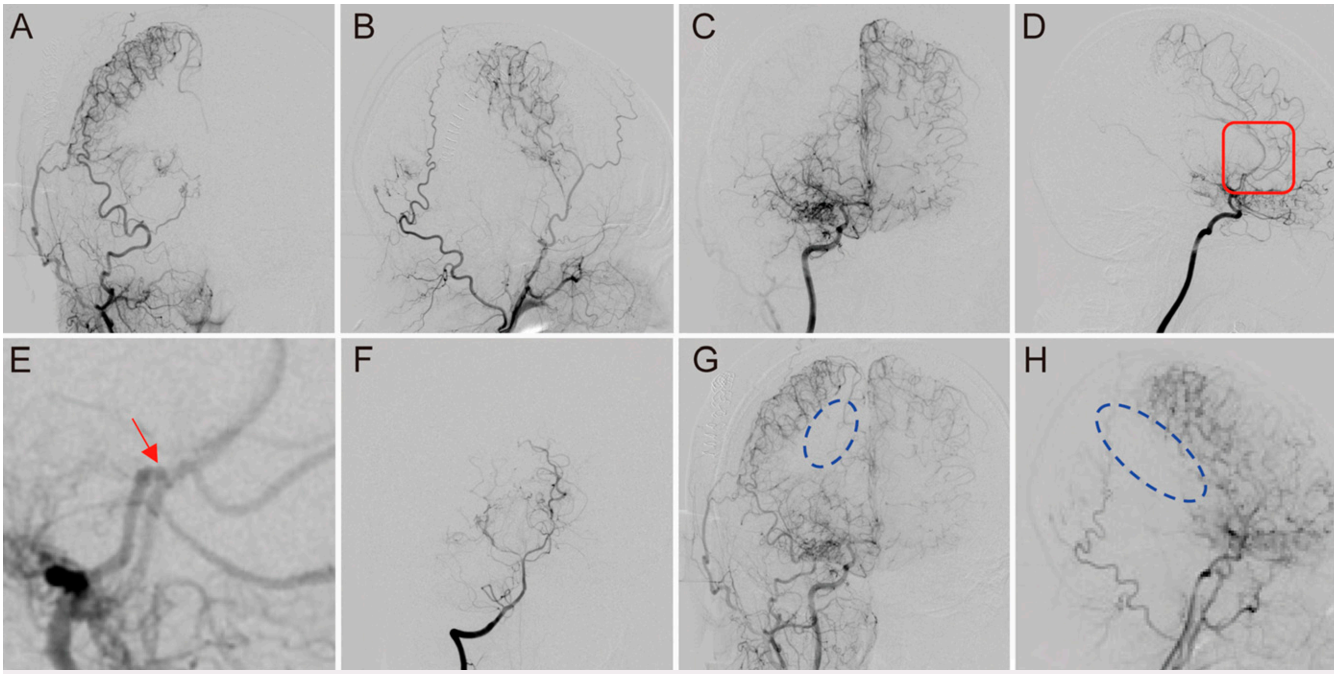

FIG. 4. Postoperative DSA. Right external carotid artery angiograms, anteroposterior view (**A**) and lateral view (**B**), showed that STA-MCA bypass covered approximately one-half of the right MCA region. Right internal carotid artery angiograms, anteroposterior view (**C**) and lateral view (**D**), revealed that the right ACA, which originally supplied blood flow to the cortical area of the ACA and MCA regions, was almost occluded at proximal A2 segment. Enlarged view (**E**) of the area enclosed by the *red square* in panel D. The blood flow of the right ACA was decreased remarkably distal to the preoperatively stenosed point at the proximal portion of A2 (*red arrow*). Right vertebral artery angiogram, anteroposterior view (**F**), showed that the perfusion area of the right PCA was not changed. Common carotid artery angiograms, anteroposterior view (**G**) and lateral view (**H**), revealed avascular areas that correspond to the right inferior parietal lobule and white matter of the right frontal lobe (*blue circles*).

anterograde flow at the stenosed point, it would be preferable to prepare the donor flow that provides sufficient perfusion distal to the stenosis, such as double STA-MCA bypass.[7]

Lessons

STA-MCA bypass for patients with MMD can cause an improper watershed shift by inducing a decrease in the original cerebral flow. Specifically, in the case of a major cerebral artery that widely supplies blood flow but has a stenosed portion, considering the occlusion risk of anterograde flow just after the stenosed point due to the reverse flow by direct bypass, preparing a donor that can supply sufficient blood flow distal to the stenosis may be preferable.

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Disclosures

Dr. Uda reported personal fees from Mizuho Corporation outside the submitted work. No other disclosures were reported.

Author Contributions

Conception and design: Kanamori, Mamiya, Takayanagi, Nishihori. Acquisition of data: Kanamori, Araki, Yokoyama, Uda, Mamiya, Nohira, Takayanagi, Ishii. Analysis and interpretation of data: Kanamori, Araki, Takayanagi. Drafting the article: Kanamori, Takayanagi. Critically revising the article: Saito. Reviewed submitted version of manuscript: Kanamori, Yokoyama, Uda, Nishihori, Izumi, Saito. Approved the final version of the manuscript on behalf of all authors: Kanamori. Study supervision: Nishihori, Saito.

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