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Case Report

Atypical postoperative radiographical findings in a patient with moyamoya disease: A case report

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ABSTRACT

Moyamoya disease is a chronic cerebrovascular disease characterized by spontaneous and progressive stenosis or occlusion of the internal carotid artery and its branches. Revascularization procedures have been shown to improve cerebral hemodynamics and decrease the risk of strokes, but several postoperative complications are known to occur.A 14-year-old girl with moyamoya disease with a history of left-sided revascularization surgery underwent right-sided revascularization. On postoperative day 4, she experienced a transient neurological event (left hemiparesis). Magnetic resonance imaging revealed large cortical and subcortical hyperintense lesions in the middle cerebral artery territory on diffusion-weighted imaging and apparent diffusion coefficient imaging. Subsequently, the radiographic findings improved within several days with resolution of the symptoms.

This case is a reminder that hemodynamic complications can develop subacutely in patients who have undergone successful revascularization for moyamoya disease. The radiological features and mechanisms of this rare condition associated with revascularization surgery for moyamoya disease are discussed.

Introduction

In patients with moyamoya disease (MMD), revascularization surgery is an established treatment to improve hemodynamic impairments and prevent ischemic or hemorrhagic strokes. [3] However, during the acute period after bypass surgery, transient neurological events and several abnormal radiological findings have been noted. Here, we present a case with a fairly rare complication with characteristic radiological findings after surgery.

Case description

A 14-year-old girl had experienced transient right hemiparesis after exercise one year prior to presentation. At that time,

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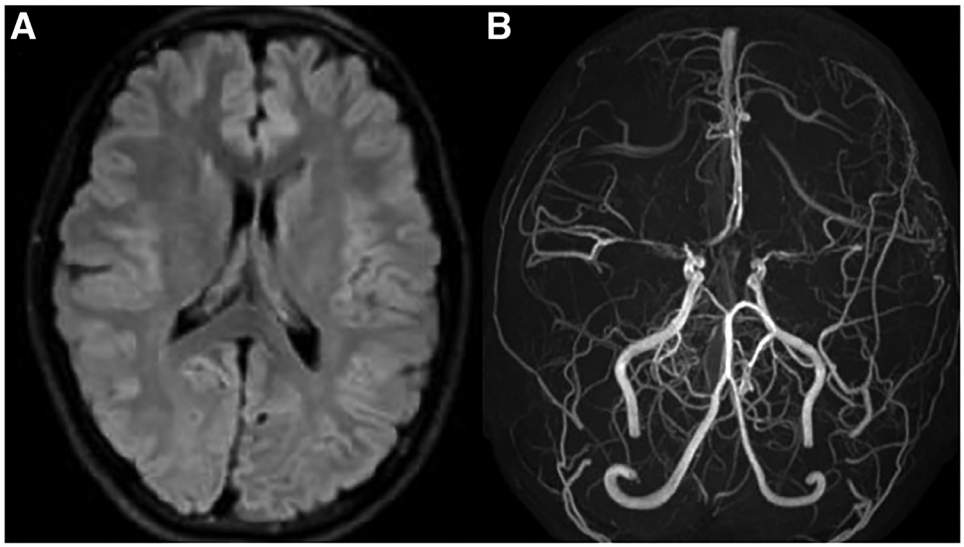

Fig. 1 – Preoperative diffusion-weighted imaging (A) revealed no ischemic lesions and magnetic resonance angiography (B) revealed the finding of MMD with no remarkable changes compared to the initial presentation one year before.

magnetic resonance imaging (MRI) and angiography (MRA) revealed MMD, and single-photon emission computed tomography (SPECT) showed marked decrease in cerebral blood flow (CBF) and cerebral vascular reserve in the left hemisphere. She underwent left superficial temporal artery (STA)-to-middle cerebral artery (MCA) anastomosis with encephalo-duro-myosynangiosis (EDMS). Her postoperative course was uneventful, and she was discharged without complications.

Although MRI and MRA revealed no new lesions (Fig. 1), she developed transient left hypoesthesia after exercise during the one-year follow-up. To evaluate her CBF, SPECT examination was performed, but the patient had an allergic reaction (thought to be due to the radiopharmaceutical agent, N-isopropyl-(123I)-p-iodoamphetamine) and the study was not successful.

She underwent right STA-MCA anastomosis and EDMS one year after the initial surgery. The procedures were uneventful and the occlusion time was 38 minutes. She recovered from the anesthesia without neurological deficit, and MRI on postoperative day (POD) 1 demonstrated no ischemic lesions and patent bypass, although swelling of the temporal muscle attached to the brain surface was noted (Fig. 2). However, on POD 4, she abruptly demonstrated left hemiparesis and an emergency MRI revealed large hyperintense and hypointense lesions in the MCA territory on diffusion-weighted imaging (DWI) and apparent diffusion coefficient(ADC) imaging, which is atypical compared to the reported postoperative findings of MMD patients (Fig. 3 A-B). We could not perform SPECT examination because of her previously mentioned allergic reaction. Instead, we tried to evaluate her CBF using MRI with arterial spin labelling, but the results obtained were poor. However, this symptom improved rapidly, and only slight hemiparesis remained on POD 5. Although DWI and ADC on POD 7 showed worse findings in comparison to POD 4 (Fig. 3 C-D), the majority of the abnormal findings had disappeared by POD 14 (Fig. 3 E-F). Finally, she was transferred to rehabilitation with modified Rankin Scale (mRS) 1 on POD 21.

At the last follow-up 3 years after the 2nd operation, the high-intensity areas of FLAIR-MRI were smaller than those on POD 14 (Fig. 3 G-H), and her left hemiparesis had resolved completely (mRS 0). The bypass patency was confirmed on MRA throughout the postoperative periods.

Discussion

MMD patients have a risk of ischemic or hemorrhagic strokes because of their intracranial artery stenosis and hemodynamic stress on vulnerable vessels. Revascularization surgery for improving a patient's hemodynamics can prevent the development of strokes, but is known to be associated with perioperative cerebral infarction and cerebral hyperperfusion causing transient neurological deterioration, delayed intracerebral hemorrhage, and vasogenic edema. [1–7] In this case, radiologic examination revealed no ischemic lesion on POD 1, but within 3 days, abnormal intensity changes were noted throughout the cortical and subcortical regions.

Notably, the patient's neurological deficit was mild and transient, even though abnormal findings on MRI were recognized in a large MCA area. Slightly delayed from neurological finding, the lesions subacutely improved. Proposed mechanisms for this complication include hyperperfusion, venous congestion due to compression by the swelling temporal muscle, and ischemic vasogenic edema. Tashiro et al. reported that vasogenic edema may be associated not only with cerebral hyperperfusion but also with hypoperfusion after STA-MCA anastomosis for MMD. [7]

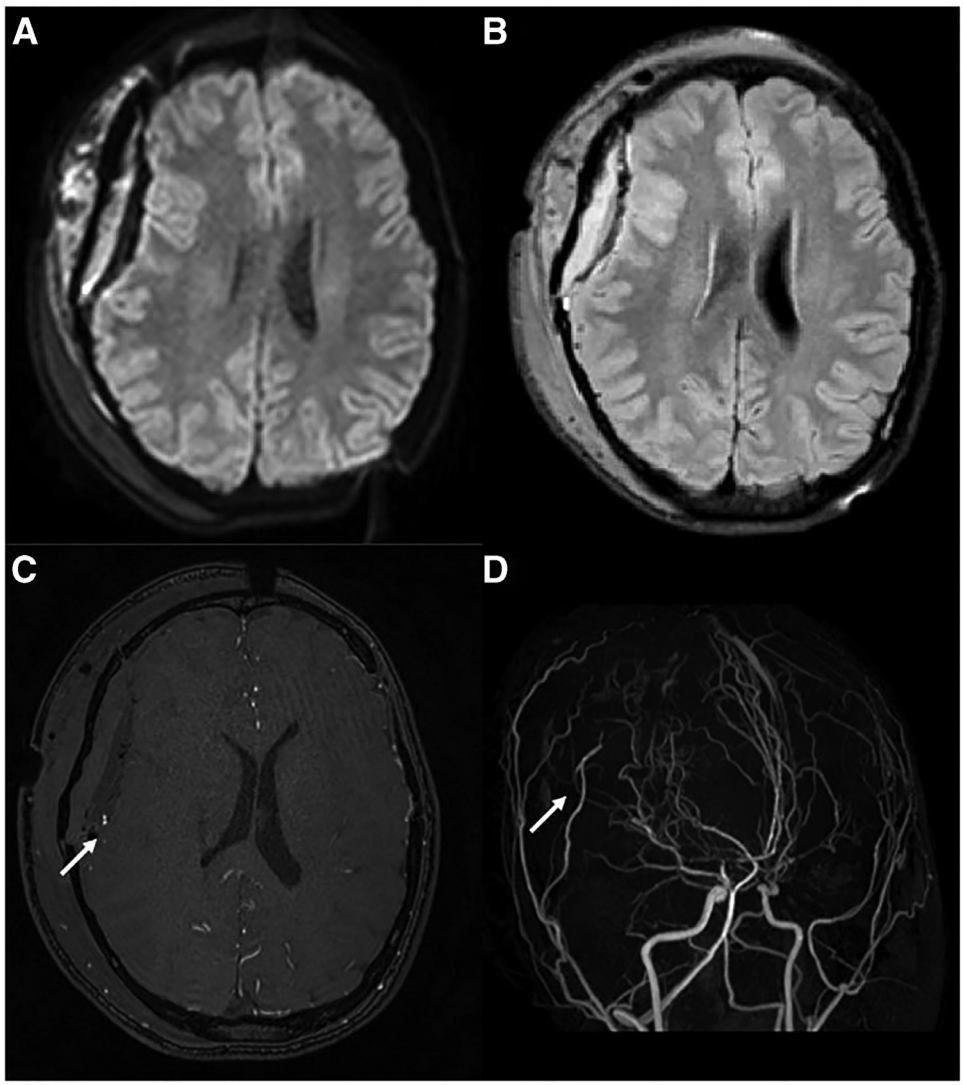

Fig. 2 – Diffusion-weighted imaging (A) and fluid-attenuated inversion recovery imaging (B) on postoperative day (POD) 1 revealed no ischemic lesions, although temporal muscle used for the indirect bypass was swelling. Magnetic resonance angiography on POD 1 (C, D) demonstrated that the bypass was patent (arrow).

Cho et al. classified infarct patterns of MMD patients on imaging into 7 groups and found that different patterns were associated with childhood-onset vs. adult-onset MMD. It was thought the age-related differences were related to differences in the vulnerability of the brain to ischemia, the stage of arteriopathy, or changes of abnormal collateral pathways. [1] Atypical postoperative radiographical findings in the present case was not in accordance with this classification and the precise etiology in our patient remains to be elucidated, especially because the postoperative CBF study could not be performed due to allergy. This case serves as a reminder that abnormal, rare radiologic findings may develop in the subacute phase after revascularization for MMD, even though no abnormal findings were recognized in the acute postoperative phase. Further study of similar cases will help to define this rare condition associated with MMD.

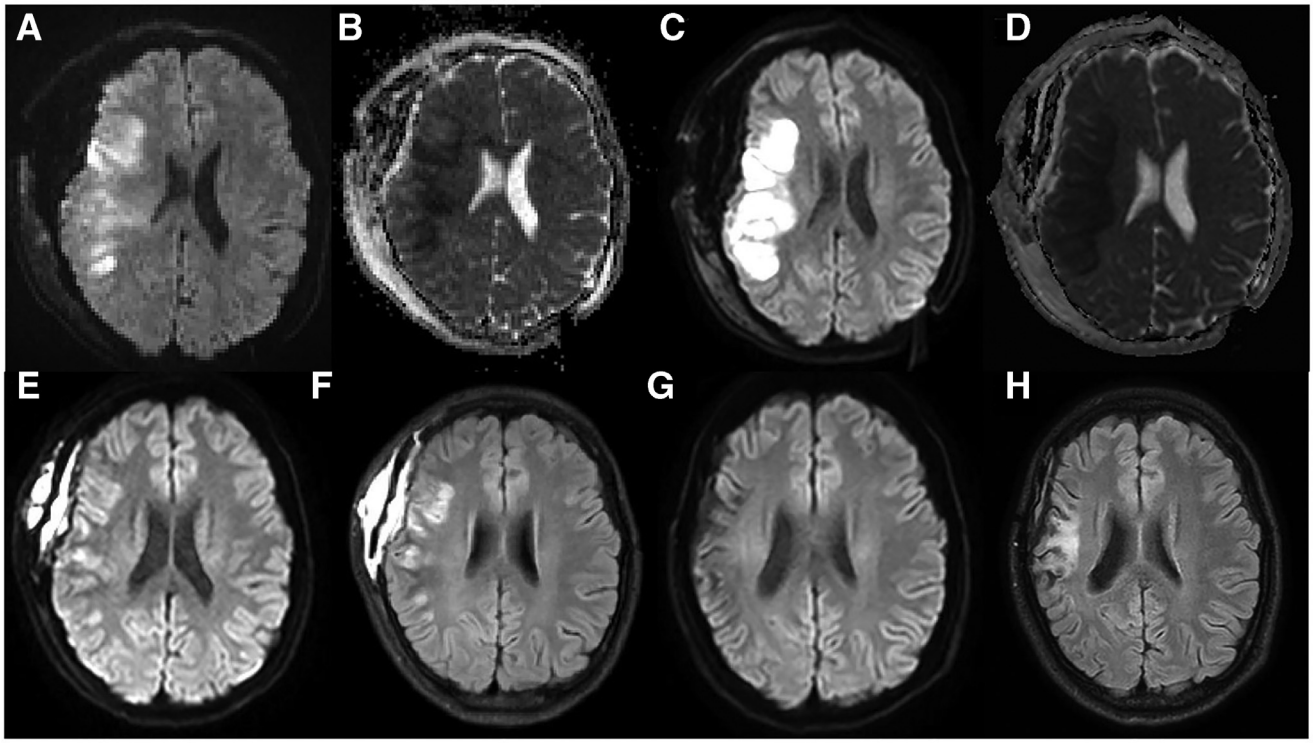

Fig. 3 – Diffusion-weighted imaging (DWI) (A) and apparent diffusion coefficient (ADC) imaging (B) on postoperative day (POD) 4 revealed large hyperintense and hypointense lesions including those in the cortical and subcortical area. DWI (C) and ADC (D) on POD 7 showed slightly worse findings compared to those on POD 4. DWI (E) and fluid attenuated inversion recovery (FLAIR) (F) on POD 14 revealed that the majority of the abnormal findings had disappeared. DWI (G) and FLAIR (H) 3 years after the 2nd operation revealed atypical lesions that were smaller compared to the findings on POD 14.

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None

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.radcr.2019.01.019.

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