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Usefulness of Three-dimensional Computer Graphics for the Direct Surgery of Ruptured Aneurysms in Deep Collateral Arteries Arising after Indirect Revascularization for Moyamoya Disease: Report of Two Cases

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Abstract

Treating ruptured aneurysms in deep collateral arteries in moyamoya disease is difficult. Two cases of intracranial hemorrhage due to ruptured aneurysms in the deep collateral vessels after indirect revascularization for moyamoya disease were treated via direct surgery with the assistance of surgical simulation using three-dimensional computer graphics. The three-dimensional computer graphics provided detailed anatomical relationships between the aneurysm and the surrounding structures, which led to successful surgical results in both patients. Ruptured aneurysms in deep collateral vessels in moyamoya disease could be successfully treated via direct surgery with the assistance of surgical simulation through three-dimensional computer graphics.

Keywords: aneurysm, moyamoya disease, simulation

Introduction

Moyamoya disease is a chronic progressive steno-occlusive disease of the distal internal carotid artery of unknown cause.[1,2] Unlike pediatric patients who present with transient ischemic attack (TIA) or stroke, approximately half of adult patients present with intracranial hemorrhage, which can have a remarkable prognostic impact. Moreover, choroidal anastomosis and lesions in the posterior cerebral artery are characteristic of posterior hemorrhage in moyamoya disease.[1,3] Most patients suffer from ischemic sequela, but some show hemorrhagic presentation.[3] A network of collateral vessels is formed bypassing the stenotic occlusion, and aneurysms tend to take place in the collateral vessels.[4] The choroidal collateral vessels are the most common site of collateral artery aneurysm and associated hemorrhage.[5] Nevertheless, direct aneurysm repair for these aneurysms had a rate of complication of up to 25%.[5] It would not be straightforward for neurosurgeons to evaluate whether a radical approach could be carried out based on conventional neuroimaging studies.

Recently, image-assisted technologies have been more and more utilized to improve safety in treating difficult neurosurgical pathologies.[6,7] They include image-guided resection of brain tumors[6] or development of postoperative dural arteriovenous fistulae by evaluating vascular structure and identifying risk factors.[7] In this report, we present two cases of intracranial hemorrhage caused by ruptured aneurysms in the deep collateral vessels after indirect revascularization for moyamoya disease, which were successfully treated with direct surgery with the aid of surgical simulation using three-dimensional computer graphics (3DCG).

Case Report 1

A 32-year-old woman presented to our hospital with a chief complaint of headache. She had a history of TIA due to moyamoya disease and had undergone bilateral encephalo-duro-arterio-myo-synangiosis (EDAMS) 20 years earlier at another institution. Upon admission, the Glasgow Coma Scale score was E3V4M6, and no motor paralysis

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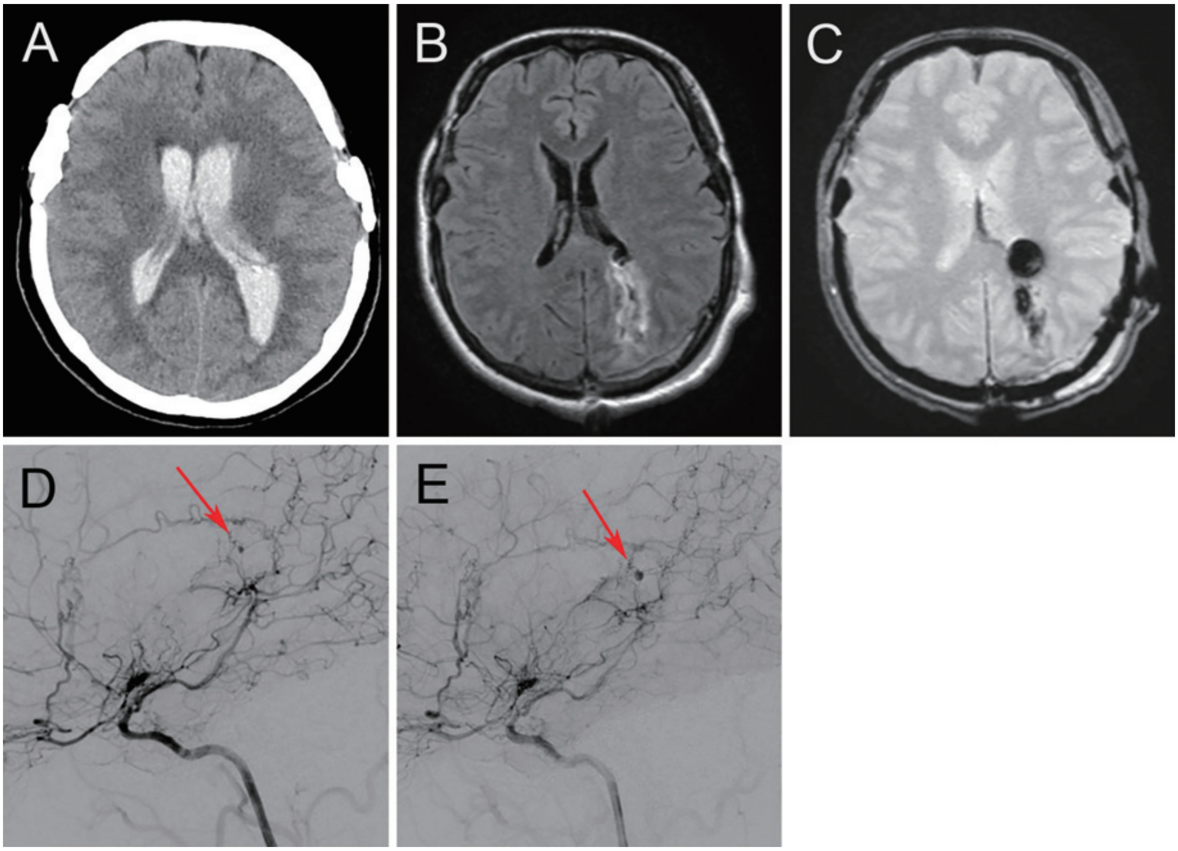

Fig. 1
Neuroimaging studies of case 1. (A) A computed tomography image showing intraventricular hemorrhage. (B) Fluid-attenuated inversion recovery and (C) T2*-weighted magnetic resonance images demonstrating mass lesion with hematoma component near the posterior horn of the left ventricle. (D) Lateral view of serial left internal carotid angiogram showing a microaneurysm at the distal part of the left anterior choroidal artery (red arrow), which grew by 1 mm 1 week later (E).

was observed in the extremities. Computed tomography (CT) images of the head revealed intraventricular hemorrhage (Fig. 1A). Fluid-attenuated inversion recovery and T2*-weighted magnetic resonance (MR) imaging presented a clot around the aneurysm (Fig. 1B-C). Angiography showed a small aneurysm at the distal portion of the left anterior choroidal artery (Fig. 1D), and no other bleeding source was suspected in other vessels. Left external carotid arteriography demonstrated a certain number of collaterals in the middle cerebral artery territory extending into the orbitofrontal and central arteries by the previous EDAMS, although the anterior parietal territory became the watershed area between the revascularization and the posterior communicating artery of the fetal type. Thus, periventricular anastomoses from the anterior choroidal artery were formed in the watershed area. Repeated angiography revealed the aneurysm growth from 2 to 3 mm in 1 week (Fig. 1E). Hence, the aneurysm was diagnosed as the source of the bleeding. Detailed anatomical relationships between the aneurysm and the surrounding structures were identified using 3DCG presurgical simulation employing the software GRID (Kompath Inc., Tokyo, Japan, Fig. 2 A-C). Arteries and the aneurysm were drawn from angiography, veins from CT images, and ventricles from MR imaging. We could recognize that part of the aneurysm protruded inside the ventricle, and the position of the parent artery was clearly confirmed, both of which were challenging to recognize via conventional imaging studies.

Direct surgery via a transcortical approach from the left occipital lobe, which is apart from previous craniotomies, was conducted, and the aneurysm was smoothly identified as simulated during the surgery. Aneurysm trapping was carried out with guidance by 3DCG simulation to confirm anatomic consistency. We also used navigation system (Fig. 2D-F). No new neurological or ischemic complication was observed, and the patient was discharged 10 days after surgery. Postoperative CT angiography revealed successful trapping and disappearance of the aneurysm (Fig. 2G-H).

Case Report 2

A 49-year-old man presented to our hospital with a complaint of headache. He also had a history of TIA attack owing to moyamoya disease and had undergone bilateral

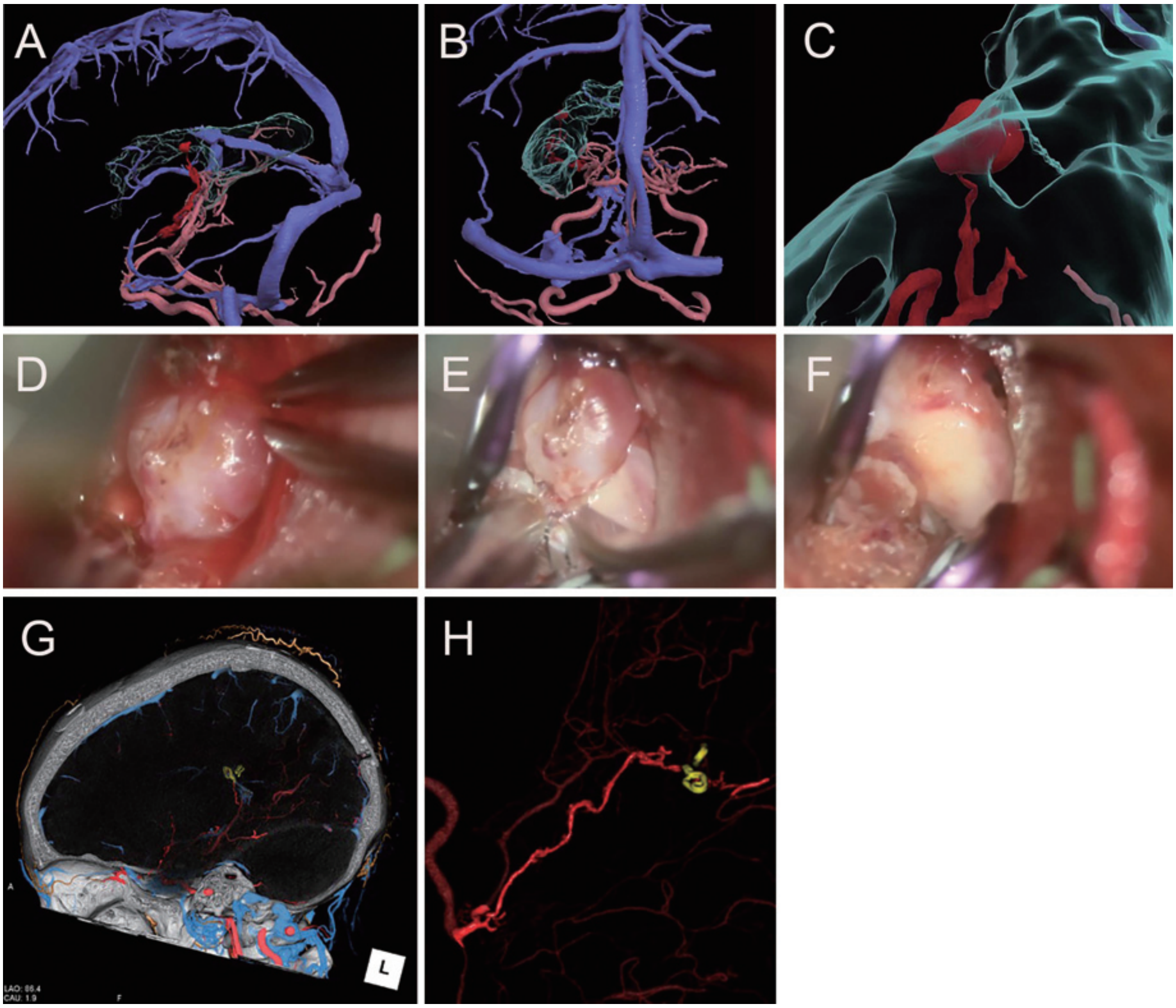

Fig. 2
(A) Lateral view, (B) posterior view, and (C) enlarged posterior view of presurgical simulation images of case 1 showing detailed anatomical relationships between the aneurysm and the surrounding structures. Red and pink: arteries; blue: veins; light blue: ventricles. As intraoperative findings, (D) An aneurysm was observed at the wall of the left lateral ventricle. (E) Aneurysm was trapped and removed (F). Postoperative computed tomography angiography (G: lateral view, H: target artery alone with same projection) presented successful trapping and disappearance of the aneurysm.

EDAMS 38 years earlier at another institution. Upon admission, he was conscious and had no neurological deficit. CT images of the head revealed hemorrhage in the left thalamus and the third ventricle (Fig. 3A). MR images presented a small mass with contrast enhancement at the edge in the left thalamus and surrounding hematoma (Fig. 3B-C). Angiography showed a small aneurysm at the left thalamoperforating artery, which was diagnosed as the source of hemorrhage (Fig. 3D). Unlike in the first case, right external carotid arteriography showed few collaterals by the previous EDAMS. Radical treatment was planned, considering that the aneurysm did not resolve on imaging follow-up (Fig. 3E). To deliberate whether open surgery or endovascular treatment would be better to eradicate the aneurysm, 3DCG simulation was employed (Fig. 4A-D). The aneurysm was located at the branching site of the left thalamoperforating artery, and its distal end was anastomosed with the medial and lateral posterior choroidal arteries. Even if the vessels that surround the aneurysm were embolized with endovascular treatment, we considered that completely occluding the parent artery would be difficult because of the anastomoses. Accordingly, we decided to conduct a transcallosal approach to treat the aneurysm to minimize brain damage. Since the massa intermedia would be an obstacle to the approach into the third ventricle, we anticipated that the massa intermedia would need to be transected intraoperatively.

During the open surgery via frontal transcallosal approach apart from previous craniotomies, the left thalamoperforating artery was followed as far as possible to the periphery, but direct observation of the aneurysm was not possible because of the limited surgical field (Fig. 4E-

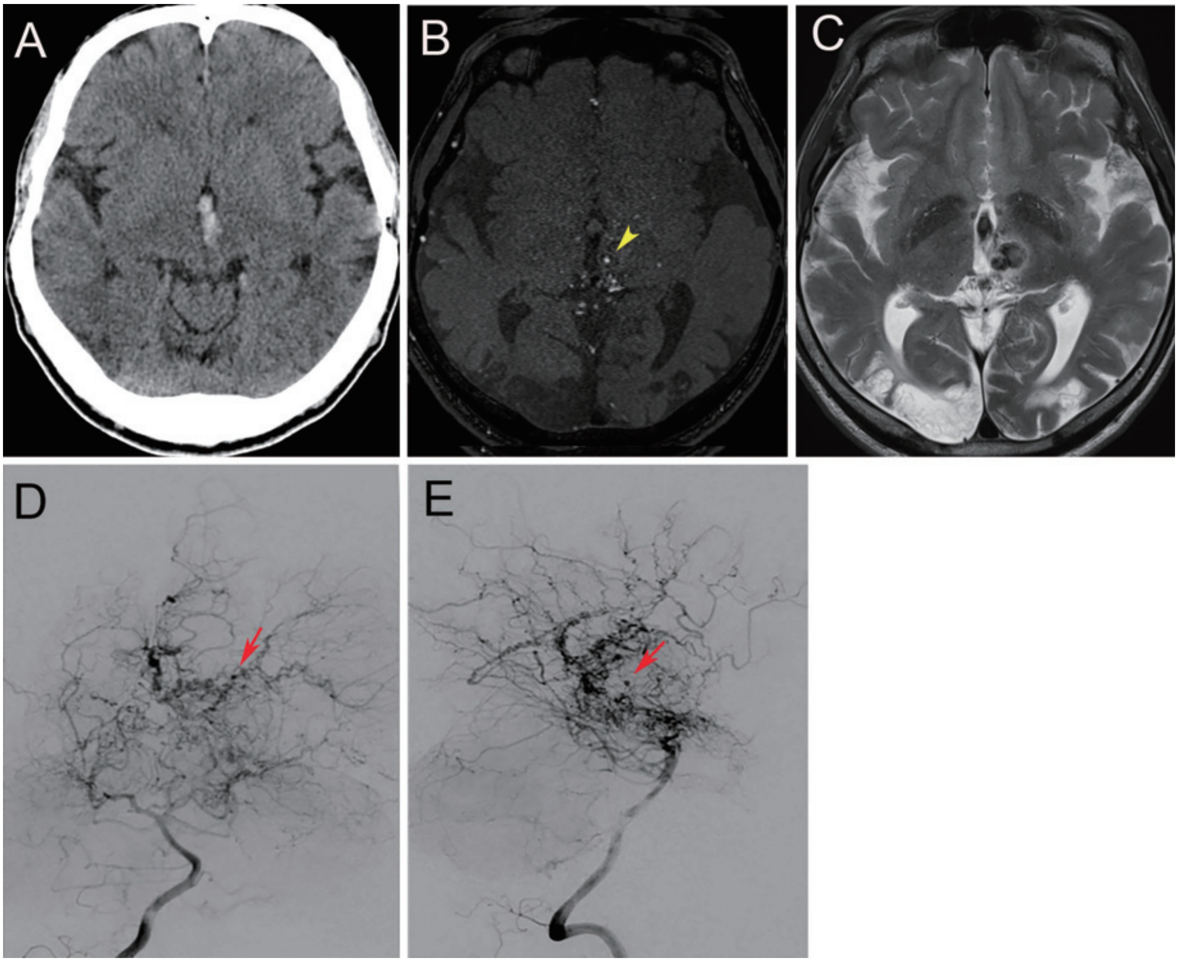

Fig. 3
Neuroimaging studies of case 2. (A) An axial computed tomography image showing hemorrhage at the third ventricle. (B) Time of flight magnetic resonance image suggesting a small aneurysm in the left thalamus (yellow arrowhead). (C) T2-weighted magnetic resonance image showing hematoma surrounding the aneurysm. Anteroposterior (D) and lateral (E) view of right vertebral arteriography showing a small aneurysm in the left thalamoperforating artery (red arrow).

F). Guided by 3DCG simulation, the thalamoperforating artery proximal to the aneurysm was occluded by an aneurysm clip. Indocyanine green angiography presented no flow even from distal to the clip. On the 3rd postoperative day, through 3D angiography, the aneurysm was confirmed to have disappeared (Fig. 4G). The patient showed transient disconnection syndrome but was discharged home after its improvement.

Discussion

We reported the successful surgical results of ruptured aneurysms in deep collateral arteries in two patients of moyamoya disease guided by 3DCG surgical simulation. Postoperative complications could be minimized through image-assisted techniques utilized for preoperative therapeutic planning and intraoperative guidance. Both our patients experienced hemorrhage 20 and 38 years after indirect bypass for ischemic moyamoya disease. Since a previous study reported hemorrhage in three patients after indirect bypass for 172 patients (1.7%),[8] hemorrhage can take place after indirect bypass. Other studies reported hemorrhage in three patients more than 7 years after direct bypass for 58 patients (5%),[9] and repeated hemorrhage after direct[10] or combined[11] revascularization for hemorrhagic moyamoya disease was also reported.

Therapeutic indication for hemorrhagic moyamoya disease

Given the treatment indication for moyamoya disease with hemorrhagic presentation, both of our patients had undergone indirect revascularization surgery, and additional management was required. If aneurysms arise from the choroidal artery, most of them are pseudoaneurysms.[4] Those with hemorrhagic presentation were more likely to deteriorate due to rebleeding,[12] which led to a high mortal-

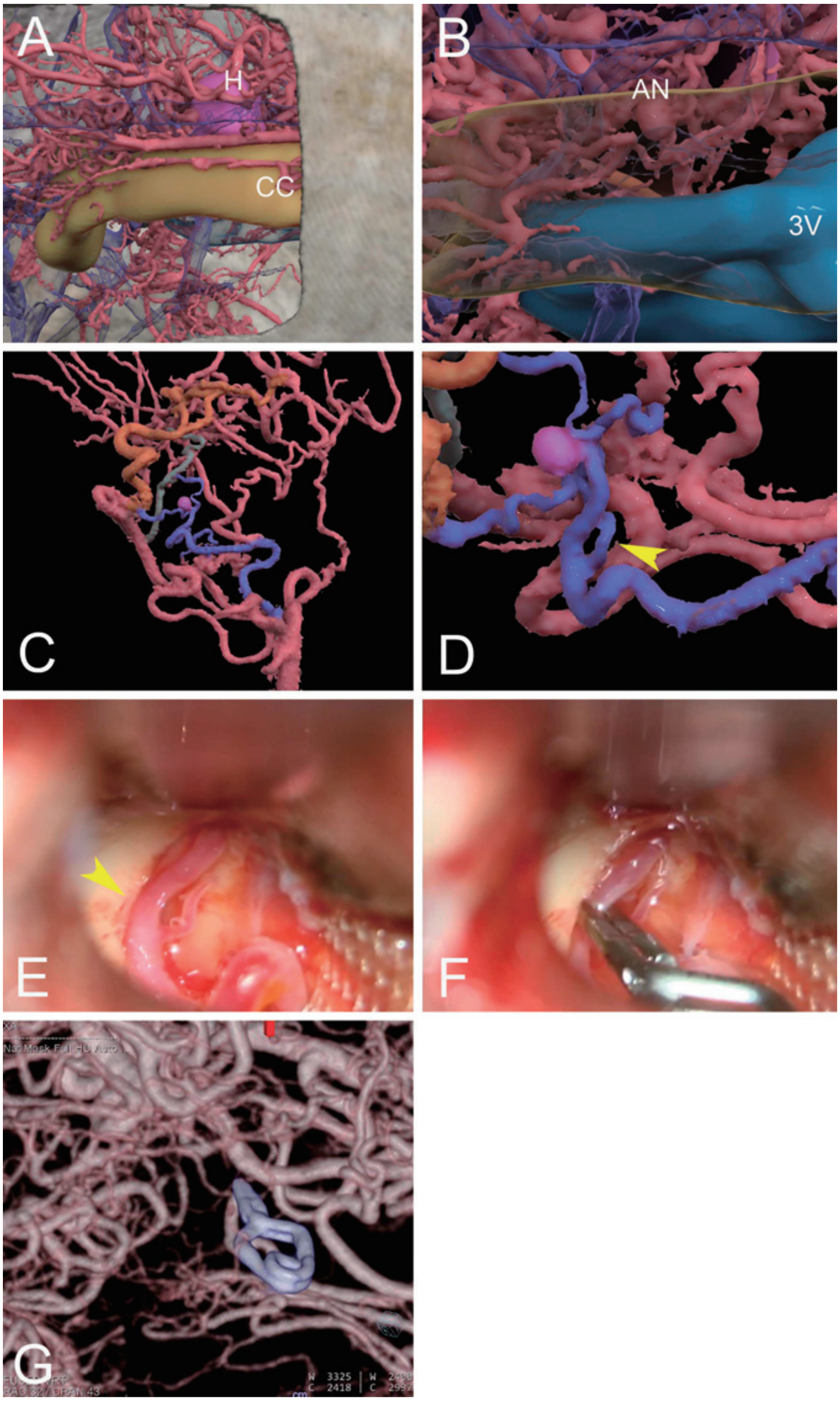

Fig. 4
(A) Presurgical simulation images of case 2 showing a posterior view to the corpus callosum (CC) near hematoma (H). (B) The aneurysm (AN) was found to be located next to the third ventricle (3V) and was found to be reachable via the corpus callosum. (C) Detailed vascular architecture around the aneurysm and its enlarged view (D) showing the left thalamoperforating artery as the parent artery of the aneurysm (yellow arrowhead).
As intraoperative findings, (E) the left thalamoperforating artery as the parent artery of the aneurysm could be observed as simulated. The position of the yellow arrowhead is identical to that in Fig. 4D. (F) An aneurysm clip was applied to the point of the yellow arrowhead. (G) Postoperative 3D angiogram confirming the disappearance of the aneurysm. Purple: aneurysm; pink: basilar artery and its branches; blue: lt thalamoperforating artery; gray: lateral posterior choroidal artery; orange: medial posterior choroidal artery.

ity. Rebleeding often takes place long after the initial onset, even 20 years later.[13] Therefore, more radical long-term preventive measures against rebleeding must be implemented.

Selection of treatment option by 3DCG

Precise and detailed surgical planning by 3DCG in our two patients enabled successful surgical results. Aneurysms relating to moyamoya disease arise in distal arteries, and most of them are fusiform or pseudoaneurysm.[7] Potential ischemic risk has to be considered in treating these aneurysms. Even when endovascular embolization is to be selected, these distal aneurysms may not be accessed due to the tortuous vessel.[5,14] Direct approach to deep aneurysms can also lead to white matter damage and ischemia of the surrounding parenchyma. 3DCG was useful in the selection of treatment modalities. The addition of direct bypass surgery might be one of the therapeutic options.[10] Nevertheless, we considered re-craniotomy to have the risk of damaging previous indirect revascularization. Since microaneurysms in both our patients could be identified as sources of bleeding by detailed imaging studies, a direct approach for aneurysms was selected.

Usefulness of 3DCG in comparison to neuronavigation

Although neuronavigation is widely employed in a large proportion of neurosurgical operations nowadays,[14] 3DCG was additionally used in our patients. In the second case, in particular, intraoperative morphological evaluation of the aneurysm embedded in the brain parenchyma and surrounding arterial pathway made it possible to obliterate the parent artery of the aneurysm without excessive invasion, which thereby eliminated postoperative severe complications. Although relatively simple surgical simulation and planning are also possible through recent neuronavigation, interactive and intuitive manipulation after segmentation of vessels and surrounding structures in detail might be only possible in our 3DCG. 3DCG was an extremely powerful tool for detailed planning of treating relatively small targets such as in our patients.

Limitations

There exist several limitations. First, the 3D images of detailed vascular structures are created using artificial intelligence (AI) based on CT, MR, and angiography imaging information. If certain structures such as small perforating arteries are difficult to visualize by AI, additional corrections have to be carried out by hand. This correction depends on the diagnostic ability of each physician. The image rendering technology for microstructures might be an issue to be addressed in the future. Second, the skill of selecting images for surgery out of a lot of imaging information would be necessary.[6] Specialized neurosurgeons who are familiar with surgical approaches must engage in creating 3DCG planning.

Conclusions

We were successful in employing the direct approach for ruptured small aneurysms in the deep collateral vessels in two patients with moyamoya disease with the assistance of 3DCG. Postoperative complications could be minimized through image-assisted techniques utilized for preoperative therapeutic planning and intraoperative reference.

Informed Consent

The patients provided informed consent for publication.

Conflicts of Interest Disclosure

The authors declare no conflicts of interest that are directly related to the content of this article.

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