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CASE REPORT

Intracerebral hemorrhage caused by rupture of a giant aneurysm complicating superficial temporal artery–middle cerebral artery anastomosis for moyamoya disease

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Abstract
Introduction Aneurysm formation at the anastomosis site after extracranial–intracranial (EC–IC) bypass surgery for major arterial occlusion or stenosis due to atherosclerosis has only been reported a few times previously. However, no case describing the formation of a giant aneurysm after EC–IC bypass surgery has been reported to date. Additionally, this complication associated with moyamoya disease is extremely rare, and only one case has been reported so far.
Clinical report We report a case of a 51-year-old woman having a rare complication of intracerebral hemorrhage due to rupture of a giant aneurysm that developed after superficial temporal artery–middle cerebral artery anastomosis for the treatment of moyamoya disease.
Conclusion To the best of our knowledge, this is the first reported case of a giant aneurysm, also the largest so far occurring after EC–IC bypass surgery and the second reported case of a rupture of an aneurysm formed after bypass surgery for moyamoya disease.

Keywords Giant aneurysm · STA–MCA anastomosis · Moyamoya disease · Intracerebral hemorrhage

Introduction

Moyamoya disease is a progressive cerebrovascular occlusive disease that causes an abnormal enlargement of the perforating arteries (moyamoya vessels) at the base of the brain [12]. Surgical revascularization in moyamoya disease prevents cerebral ischemic attacks by improving the cerebral blood flow (CBF). Superficial temporal artery–middle cerebral artery (STA–MCA) anastomosis with or without indirect bypass is generally used as the standard surgical treatment of moyamoya disease [2, 3, 10]. However, various complications can occur after STA–MCA anastomosis for moyamoya disease, such as graft occlusion, wound infection, infarction, and postoperative intracerebral hemorrhage (ICH) remote from the site of the operation [6]. ICH occurring due to the rupture of an aneurysm that has developed at the anastomosis site is an extremely rare complication, and only one case has been reported to date [1]. Although few cases have been reported in which the aneurysm formed at or close to the anastomosis site after bypass surgery for ischemic disease [4, 8, 9, 11], the size of the aneurysm has been reported to range from 4 to 13 mm, and no case of formation of a giant aneurysm after STA–MCA anastomosis has been reported to date. Here, we describe the case of a 51-year-old woman having an extremely rare complication of ICH due to the rupture of a giant aneurysm which formed at the site of anastomosis 6 months after STA–MCA anastomosis for the treatment of moyamoya disease.

Clinical report

In May 2008, a 51-year-old woman was admitted with a history of 3 years of repeated transient motor weakness in the left arm and leg, which occurred one to three times per month and always lasted for several minutes. Carotid angiography revealed occlusion at the terminal portion of the right internal cerebral artery (ICA) and at the beginning of the left M1 portion along with an abnormal basal vascular

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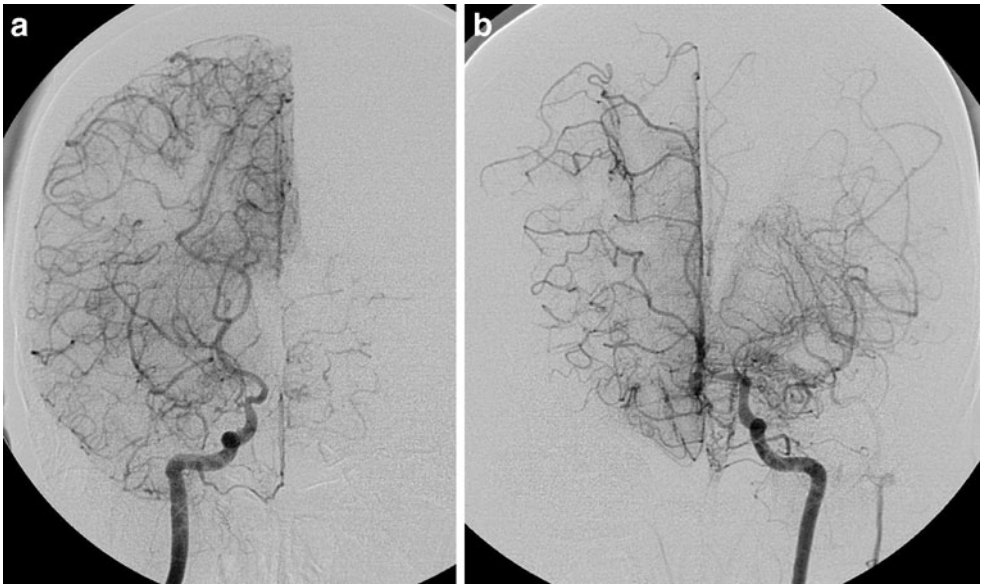

Fig. 1 **a** Right and **b** left anteroposterior carotid angiogram showing occlusion of the right supraclinoid ICA and the left M1 portion along with an abnormal basal vascular network typical of moyamoya disease

network typical of moyamoya disease (Fig. 1). Technetium-99m-hexamethyl-propyleneamineoxime (HMPAO) single-photon emission computed tomography (SPECT) with acetazolamide challenge revealed that perfusion and vascular reserve were significantly decreased in the left cerebral hemisphere. She underwent STA–MCA anastomosis and encephalomyosynangiosis (EMS) on the left side. The postoperative course was uneventful. Postoperative cerebral angiography showed a good patency of the direct bypass and collateral vasculature into the left MCA and anterior cerebral artery (ACA) territory from the left STA (Fig. 2). In addition, increased perfusion and improved vascular reserve in the left cerebral hemisphere were evident on SPECT with acetazolamide challenge 3 weeks after operation. Six months later, the patient was admitted to the emergency room at 2 a.m. with stupor and right hemiparesis after a generalized tonic–clonic convulsion. Cranial computerized tomography (CT) scans revealed a massive ICH in the left frontotemporoparietal region, which was associated with a 2.6×2.5-cm round, aneurysm-like lesion that was contiguous to the site of left temporal craniotomy (Fig. 3a). Due to a malfunction in the CT angiography program, we were unable to perform CT angiography. Because of the patient's serious condition (she was in a state of mental stupor) and the marked mass effect of hematoma in the CT image, we performed emergency evacuation of the hematoma using a frameless navigation

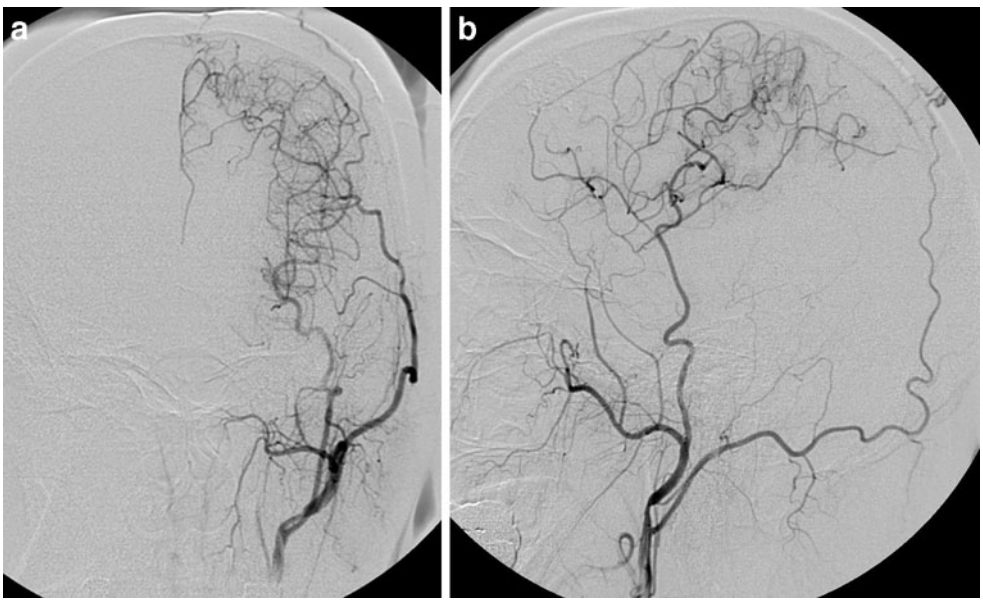

Fig. 2 **a** Anteroposterior and **b** lateral carotid angiography obtained 3 weeks after STA–MCA anastomosis showing a good patency of the direct bypass and collateral vasculature into the left MCA and ACA territory from the left STA

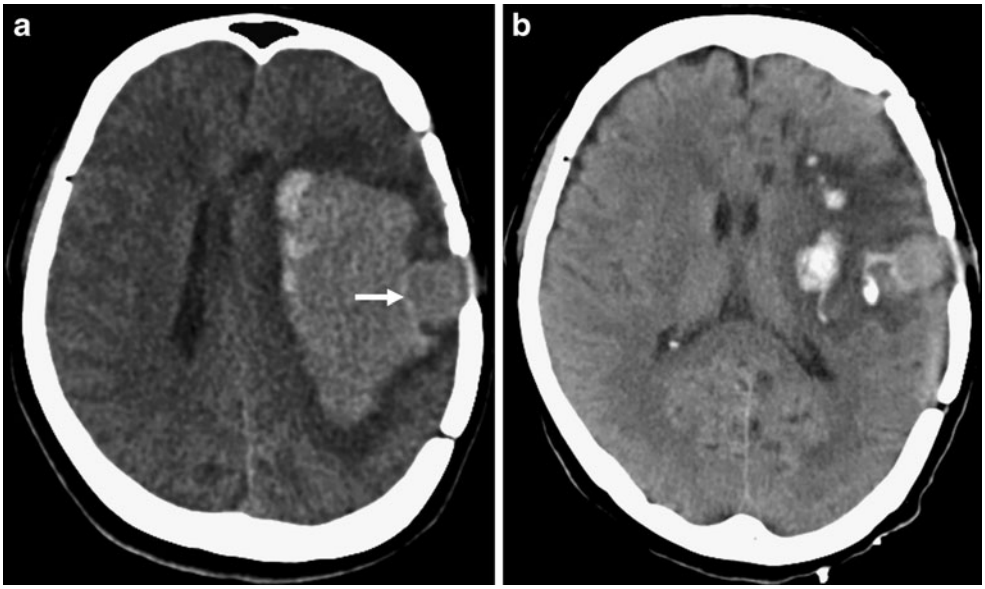

Fig. 3 **a** Cranial CT scans showing a massive ICH in the left frontotemporoparietal region, which is associated with a round, aneurysm-like lesion (*arrow*) that is contiguous to the site of left temporal craniotomy. **b** Postoperative CT scans obtained 2 days later showing almost complete removal of the hematoma except for the aneurysm-like lesion

system through a burr hole under local anesthesia. The catheter was inserted around the aneurysm-like lesion. The postoperative CT scans obtained 2 days later revealed almost complete removal of the hematoma except for the aneurysm-like lesion (Fig. 3b), with improvement in her level of consciousness. Follow-up angiography revealed a giant aneurysm at the site of STA–MCA anastomosis (Fig. 4). There was no vasculature from the aneurysm into the MCA territory. We recommended an operation, but the patient's family rejected the recommendation due to financial constraints and other factors. We finally convinced her family, and 2 months later, we performed a total aneurysmectomy through the site of the previous craniotomy. The distal STA was clipped near the site of aneurysm. It was difficult to find the recipient artery of MCA because the dura mater and layer of temporalis muscle on the cortical surface were tightly adhered to each other because of previously conducted EMS. After circumferential dissection of the aneurysm, the distal end of the STA was clipped, and en bloc resection of the previous anastomosis site including the ruptured aneurysm was then performed. The aneurysm wall protruded partially into the hematoma cavity (Fig. 5a). Histological examination of the ruptured aneurysm revealed the features of a true aneurysm (Fig. 5b). The postoperative course was uneventful,

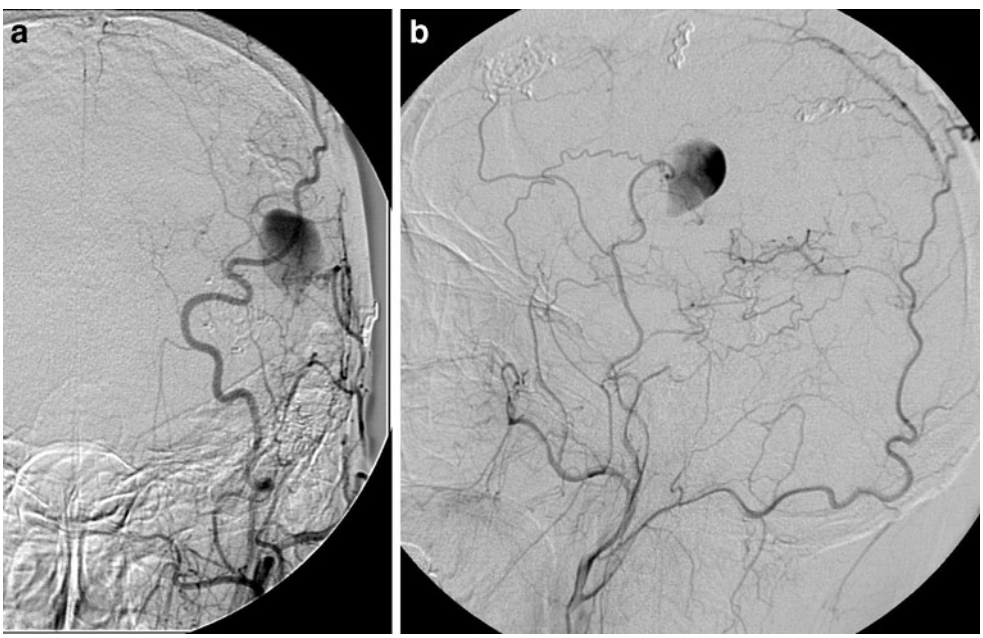

Fig. 4 **a** Anteroposterior and **b** lateral carotid angiography obtained 6 months after STA–MCA anastomosis showing a giant aneurysm of the distal STA at the site of the STA–MCA anastomosis

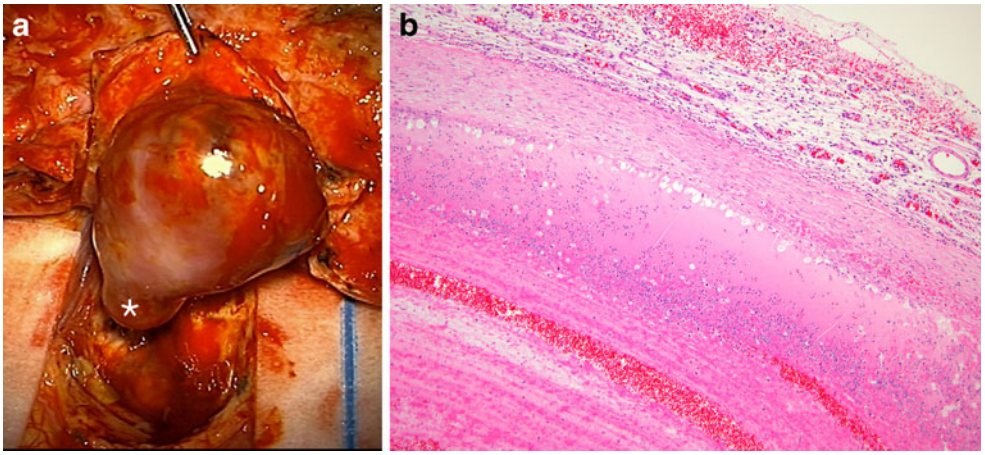

Fig. 5 **a** Intraoperative photographs showing a 26×25-mm aneurysm at the site of the STA–MCA anastomosis after circumferential dissection. Note the aneurysm wall protruding partially into the hematoma cavity (*asterisk*). **b** Histological examination of the ruptured aneurysm showing elastic and muscle fibers in the aneurysm wall, confirming the diagnosis of true aneurysm (hematoxylin and eosin stain, original magnification ×100)

and the patient was transferred to a rehabilitative care unit for the treatment of right hemiparesis.

Discussion

Extracranial–intracranial (EC–IC) bypass surgery, including STA–MCA anastomosis, was developed as a treatment for patients with ischemic cerebrovascular disease secondary to vascular lesions that were not directly repairable. The direct bypass surgery targeting the MCA has been reported to improve compromised CBF, reduce ischemic attacks, and produce sufficient and good long-term results in most cases [7]. There are several potential complications of STA–MCA anastomosis. In addition to perioperative cerebral infarct or ICH after EC–IC bypass surgery, the development of an aneurysm at or close to the anastomotic site has also been reported as a major complication in a small number of patients [4, 8, 9, 11]. Although the underlying mechanism of the formation of an aneurysm after STA–MCA anastomosis remains unclear, several possible mechanisms have been suggested. One possible mechanism is a structural change in the arterial wall, such as an atherosclerotic change. Kohno et al. [4] hypothesized that an excessive jet flow from the enlarged STA impinged on the artificial bifurcation made by the STA–MCA anastomosis, resulting in the fusiform-shaped enlargement at the anastomosis site and subsequent aneurysmal dilatation caused by increased hemodynamic forces. Another possible mechanism could be the induction of a newly formed traumatic aneurysm due to intraoperative disruption of the internal elastic lamina and media of the anastomosed arteries. An immediate increase in the MCA blood pressure and blood flow may also contribute to its development [4, 11]. Vascular injury by temporary clipping or excessive adventitial stripping could be another causative factor [5].

Several groups have reported aneurysm formation at the anastomosis site after EC–IC bypass surgery for major arterial occlusion or stenosis due to atherosclerosis. In contrast, this complication associated with moyamoya disease has rarely been reported, and only one case has been described in literature [8]. In the ICH patient reported by Nishimoto et al. [8], 20 years had passed between bypass surgery and the development of ICH due to aneurysm rupture. In contrast, our patient developed ICH within 6 months of the bypass surgery. On the basis of the very long duration, Nishimoto et al. [8] speculated that a gradual increase in hemodynamic stress over a period of 20 years due to moyamoya disease, which might have caused the formation and rupture of the aneurysm at the site of anastomosis, has a pathogenesis different from that of atherosclerotic occlusive disease. Our patient had a giant aneurysm that suddenly and within 6 months grew larger (26 mm in diameter, at maximum) and ruptured. Thus, we thought that structural changes and intraoperative disruption of arteries, mentioned above as two possible mechanisms of the formation of an aneurysm after anastomosis, functioned together to cause the giant aneurysm in our patient. Further, it is assumed that the temporalis muscle around the anastomosis site effectively protected the aneurysm and enabled it to grow to its giant size.

Conclusion

To the best of our knowledge, this is the first case of a giant aneurysm and also the largest that occurred after EC–IC bypass surgery. In addition, it is only the second case of an aneurysm formed after bypass surgery for moyamoya disease. Although patients do not exhibit any specific symptoms or signs after bypass surgery, neurosurgeons should always consider the possibility of an aneurysm and should regularly conduct magnetic resonance angiography or CT angiography to check for their presence; this would help prevent serious complications such as ICH in these patients.

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Comments

The authors present the first case of a ruptured giant aneurysm at a bypass site in moyamoya disease. As the authors speculate, "structural changes and intraoperative disruption of arteries" may have played a significant role in causing the aneurysm in addition to hemodynamic changes and perhaps pathological walls of arteries in moyamoya disease per se. Quite surprisingly, the aneurysm developed in only 6 months unlike in the other moyamoya patient described by others in whom it took years to develop a nongiant aneurysm. Based on these anecdotal cases, it is difficult to give recommendations on radiological follow-up. However, patients with moyamoya are usually followed-up on regular basis anyway, and most patients necessitating any surgical procedures (obviously, in most cases revascularization) are identified. In bypass patients in general, the follow-up should also extend over years to exclude occlusion of the craft and at the same time possible aneurysms may be detected.

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Although management of the ruptured peripheral giant aneurysm at the site of STA–MCA anastomosis could have been done more appropriately in terms of early diagnosis (use of CT angiography) and timing of aneurysmectomy (simultaneously at the time of hematoma removal), the authors have successfully documented this quite rare complication of bypass surgery and that of moyamoya disease. Surely some hemodynamic factors at the site of anastomosis must have played a cardinal role and resulted in vascular wall changes such as wall disruption or dissection which might have took place for the formation of this giant peripheral aneurysm. We have to be aware of the importance of regular check also from the viewpoint of aneurysm formation. If we can detect it prior to its rupture, we would be able to do aneurysmectomy and simultaneously also maintain cerebral blood flow by doing microvascular reconstruction [1].

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