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Case Report

Embolization with NBCA for Ruptured Aneurysm Located in the Moyamoya-like Collateral Network Associated with Isolated Middle Cerebral Artery Occlusion

Abstract

Collateral vessel formation in moyamoya disease is a well-described phenomenon. However, the occurrence of unusual anastomosis pattern (moyamoya-like) associated with isolated middle cerebral artery (MCA) stenosis or occlusion has been reported very rarely and is not well known the relationship with aneurysm. We report a case of ruptured aneurysm treated with N-butyl cyanoacrylate (NBCA) located in moyamoya like collateral network with isolated MCA occlusion.

Keywords: *Aneurysm, moyamoya, N-butyl cyanoacrylate*

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Introduction

Atherosclerotic steno-occlusive disease of major intracranial arteries has been known the main cause of stroke worldwide and more frequent and severe than extracranial disease. In severe stenosis or occlusion of major artery with poor perfusion state, various vascular networks can be occurred. The moyamoya angiographic pattern has also been seen in association with stenosis in the posterior circulation and in association with a variety of vascular anomalies.[1] The occurrence of moyamoya like collateralization in isolated middle cerebral atherosclerotic stenosis or occlusion has been reported very rarely, and abnormal vascular network may be associated with aneurysm formation.[2] We present clinical case of ruptured aneurysms located in the collateral network of moyamoya pattern in the middle cerebral artery (MCA) occlusions.

Case Report

A 54-year-old woman with a history of hypertension was admitted to our hospital with a sudden, thunderclap headache. Neurological examination showed drowsy consciousness without language and motor impairment. Computerized tomography (CT) scan revealed subarachnoid hemorrhage in both sylvian fissure and basal cistern [Figure 1a]. CT angiography showed left proximal M1 segment occlusion with faint distal filling. There is no definite aneurysmal sac in CT angiography [Figure 1b]. Conventional and 3D rotational angiogram defined a moyamoya-like collateral network in occluded MCA area and a small aneurysm suspected as pseudoaneurysm was found in internal carotid artery (ICA) bifurcation area [Figure 1c and d]. At first, we thought ICA bifurcation aneurysm, but the selection of microcatheter and microwire into aneurysm was not achieved despite several attempts. Looking closely at angiography, we thought an aneurysm could arise from collateral artery behind ICA bifurcation. Microcatheter was navigated collateral artery, and on microcatheter angiogram, a small aneurysm located in collateral network was found [Figure 1e]. Our initial plan was to reach as close to the aneurysm as possible and embolize the aneurysm with coils or embolic material. However, accessing the microcatheter close to the aneurysm was a very demanding task because of the complex vascular network. Although attempting coiling after approaching the parent artery as close as possible to the aneurysm, the microcatheter could not provide sufficient support for coiling. We tried to embolize with lower concentration (about 30%) of NBCA glue to reach the pseudoaneurysm. Embolization with NBCA was successful, and aneurysm was not visible in angiography [Figure 1f]. Because the pseudoaneurysm was likely to

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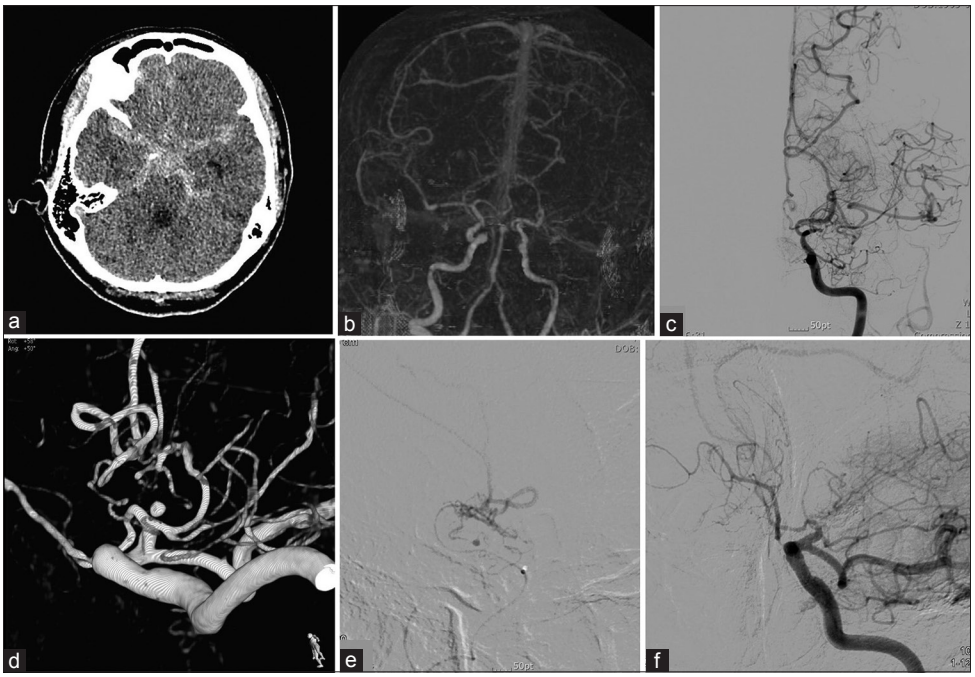

Figure 1: (a) Computerized tomography scan showed subarachnoid hemorrhage in both Sylvian fissure and basal cistern. (b) Computerized tomography angiography showed left proximal M1 segment occlusion with faint distal filling. There is no definite aneurysmal sac in computerized tomography angiography. (c) Conventional angiogram revealed a moyamoya like a collateral network in occluded middle cerebral artery area. (d) Three-dimensional rotational angiogram revealed small aneurysm (suspected as pseudoaneurysm) in internal carotid artery bifurcation area. (e) On microcatheter angiogram, a small aneurysm located in collateral network was found. (f) Embolization with N-butyl cyanoacrylate was successful, and aneurysm was not visible in angiography

be refilled due to retrograde flow within collateral network, repeated angiography with regular interval should be needed after embolization. Although at first, we intended to use coils, NBCA glue can be a handy embolic agent when the microcatheter is unable to reach to the pseudoaneurysm or could not provide enough supporting in coiling. Ten days later, follow-up angiography was performed, and there was no recanalization of aneurysm. The patient was discharged without neurologic deficit.

Discussion

A moyamoya collateral pattern in isolated MCA occlusions has also been described among Japanese patients, but there are few reports in non-Japanese patients.[3,4] However, natural history of this entity has not been well described. Most patients with isolated MCA steno-occlusive disease do not have severe perfusion impairment unless they have another intracranial vascular occlusion. Kato *et al*.[5] by PET examinations, reported significant cerebral perfusion impairment in patients with occlusion of the MCA with moyamoya phenomenon compared with patients without moyamoya vessels. In addition, Tanaka *et al*.[6] demonstrated that patients with occluded MCA and moyamoya vessels had a poor leptomeningeal collateral network. These findings suggest that patients with a poor collateral system cause significant cerebral perfusion impairment, and this may be involved in the origin of the moyamoya phenomenon. At present, various classification systems have been reported for moyamoya disease (MMD) or moyamoya syndrome (MMS) associated with intracranial aneurysm in the literature. The possible link between atherosclerotic intracranial steno-occlusive disease and aneurysmal development could be inflammation and flow disturbances, well-known pathogenic factors involved in both diseases. For example, intracranial aneurysm can be divided into pseudoaneurysm, saccular (true) aneurysm, and dissecting aneurysm, according to the morphology and histology of the aneurysm.[7,8] Alternatively, Kodama *et al*.[9] provided the classification of aneurysms arising from the circle of Willis and those from the abnormal vessel network. In MMD, an intracranial aneurysm and pseudoaneurysm are frequently present in the main and collateral vessels, and the distal aneurysm is thought to be responsible for rebleeding.[10-12] Aneurysms from the distal segments of the peripheral, cerebral arteries, or moyamoya vessels are pseudoaneurysms that come from dissection rupture or are a result of moyamoya vessel rupture. These aneurysms are formed due to increased stress on the vessel wall from the high flow imposed by occlusion of the anterior circulation.[7-9,13] These aneurysms could produce parenchymal, subarachnoid, or intraventricular hemorrhage. Treatment strategies for intracranial aneurysms associated with moyamoya pattern collateral network include surgical clipping and endovascular embolization.[14,15] Considerable dysplastic blood vessels and stiff arteries without needed plasticity can cause difficulty for the localization and exposure of aneurysms in direct surgery. Furthermore,

accurate localization of pseudoaneurysms from the distal segments of the peripheral cerebral arteries or moyamoya vessels is also challenging during surgical clipping procedures. For this reason, endovascular therapy is preferred, and different strategies can be used, such as coil or glue embolization and occlusion of the parent vessel.[10,11] The approach of the microcatheter to distal collateral vessel is extremely difficult work due to complex vascular network. In these situations, the microcatheter often may not be able to provide enough support for coiling. Embolic materials such as NBCA can make the embolization of distal aneurysms much easier.

Conclusions

Moyamoya like collateral network can occur in MCA occlusion (preferably with hypoperfusion) and can be associate with intracranial aneurysms or pseudoaneurysms. These aneurysms can cause hemorrhagic presentations; therefore, endovascular treatment (aneurysm obliteration and/or parent artery occlusion) with NBCA can be good treatment option for in complex vascular anatomy state.

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Conflicts of interest

There are no conflicts of interest

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