Middle Cerebral Artery Occlusion with Moyamoya-Like Vessels and Aneurysms

A Report of Two Cases

RODRIGO RIVERA, JUAN SORDO, LAUTARO BADILLA, EDUARDO BRAVO, RODRIGO RIVEROS, PABLO GIACAMAN

Neuroradiology Department, Alfonso Asenjo Institute of Neurosurgery; Santiago, Chile

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Summary

We describe two cases of aneurysmal rupture in moyamoya-like vessels in middle cerebral artery occlusion. This phenomenon was previously described in severe steno-occlusive disease and accounts for the hemorrhagic presentation. To our knowledge, these are the second and third clinical cases published in modern neuro-radiological literature.

Introduction

Atherosclerotic steno-occlusive disease of major intracranial arteries is the main cause of stroke worldwide, being more frequent and severe than extracranial disease¹. A moyamoya collateralization pattern can occur in isolated middle cerebral atherosclerotic stenosis² in response to poor perfusion³. Previously, only one clinical case was published, with ruptured aneurysm located in the collateral network in patients with moyamoya phenomenon secondary to middle cerebral artery occlusion. Our purpose is to describe two clinical cases of ruptured aneurysms located in the collateral network of moyamoya phenomenon in middle cerebral artery occlusions.

Clinical Cases

Case 1

An 83-year-old Hispanic woman with a history of hypertension and coronary artery disease was admitted to our hospital with an acute

thunderclap headache. Neurological examination showed impairment of consciousness and meningism.

CT scan revealed subarachnoid hemorrhage in the right sylvian fissure and hemoventricle with secondary hydrocephalus. CT angiography showed proximal right M1 segment occlusion with distal irrigation from collaterals and a post-occlusive aneurysmal dilatation.

Conventional angiogram defined a collateral pial network from the right pericallosal and right posterior cerebral arteries and revealed a 2 × 1 mm microaneurysm in the right distal M1. We opted for conservative management. The patient did not rebleed and had a good clinical course.

Case 2

A 60-year-old Hispanic man with hypertension and rheumatic disease was admitted with sudden headache. Neurological examination revealed dysarthria and left hemiparesis.

CT and CT angiography showed right ganglionic acute hemorrhage and right M1 occlusion with development of a collateral network and distal collateral aneurysmal dilatation near the bleeding site. Conventional angiography showed occlusive arteriopathy in the right middle cerebral artery, with lenticular and M2 rete mirabile and collateral vessels from anterior and posterior cerebral arteries. A 2 mm microaneurysm was found in the right lenticular rete mirabile. An intracranial bypass and microaneurysmal clipping were performed with a good outcome.

Figure 1 A) CT scan shows subarachnoid hemorrhage predominant in the right sylvian fissure, hemiventricle and hydrocephalus. B) CT angiography with occlusion of proximal right M1. C,D) Catheter angiography reveals occlusion of the right M1 segment and moyamoya-like vessels. E) 3D rotational catheter angiography shows a distal right M1 pseudoaneurysm.

Search strategy

We performed a bibliographic search on Pubmed-Medline, using the key words “moyamoya-like”, “moya moya-like”, microaneurysm, pseudoaneurysm, rete, “rete mirabile”, “moyamoya disease” [Mesh], “cerebral hemorrhage” [Mesh], “subarachnoid hemorrhage” [Mesh], “arterial occlusive diseases” [Mesh], “middle cerebral artery” [Mesh], “intracranial aneurysm” [Mesh], alone and associated with the Boolean operator “AND”. Secondly we searched in related articles of publications of interest.

Discussion

A moyamoya collateral pattern has been reported isolated in middle cerebral artery (MCA) occlusions in Japanese and non-Japa-

nese patients^{2,4-7}. Most patients with isolated MCA steno-occlusive disease do not have severe perfusion impairment unless they have another intracranial vascular occlusion^{8,9}. Moreover, Kato et al.³, by PET examinations, showed significant perfusion impairment in patients with occlusion of the middle cerebral artery and moyamoya phenomenon compared with patients without moyamoya vessels. In addition, Tanaka et al.⁸ 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 have hypoperfusion and this may be involved in the origin of the moyamoya phenomenon.

The possible link between atherosclerotic intracranial steno-occlusive disease and aneurysmal development could be inflammation (lower case) and flow disturbances, well known

Figure 2 A) CT scan with right ganglionic hemorrhage. B) CT angiography with occlusion of the proximal right M1, moyamoya-like vessels and distal aneurysmal dilatation near the hematoma. C,D) Catheter angiography reveals occlusion of the right M1 segment, moyamoya-like vessels and distal microaneurysm. E) 3D rotational catheter angiography shows a distal right lenticular microaneurysm.

pathogenic factors involved in both diseases. Intracranial aneurysms and pseudoaneurysms of major and collateral vessels are described in moyamoya disease^{10,11}, with distal collateral aneurysms thought to be responsible for rebleeding¹². Few cases of aneurysms have been reported in atypical cases (moyamoya-like) in MCA occlusion^{4-6,13}, with only one clinical case of a collateral network ruptured aneurysm⁵ to our knowledge. These aneurysms could produce parenchymal, subarachnoid or intraventricular hemorrhage⁵.

Endovascular therapy is preferred and different strategies can be used, like coil or glue embolization and occlusion of the parent vessel^{10,11}. In our cases, diagnosis was suspected with CTA but confirmed with DSA and 3D DSA that allowed an acute definition of the angioarchitecture.

In summary, moyamoya phenomenon can occur in MCA occlusion (preferably with hypoperfusion); thus, aneurysms and pseudoaneurysms can appear and they should be taken into account in hemorrhagic presentations.

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Rodrigo Rivera, MD
Neuroradiology Department
Instituto de Neurocirugía Dr Alfonso Asenjo
José Miguel Infante 553
7500691 Providencia
Santiago, Chile
E-mail: riveranrx@gmail.com