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Article

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Moyamoya Syndrome with contra lateral DACA aneurysm: First Case report with review of literature

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Abstract: Moyamoya disease is a progressive steno-occlusive disease of bilateral internal carotid artery with the development of fine collateral vessels and is an angiographic diagnosis. Concurrent Moyamoya disease with intracranial aneurysm had been reported in literature by various authors. Moyamoya disease is reported with aneurysm at various locations including basilar tip, anterior choroidal artery, posterior choroidal artery, anterior cerebral artery and middle cerebral artery. After reviewing literature thoroughly there is not a single case report on Moyamoya disease with isolated distal anterior cerebral artery aneurysm with the best of our knowledge. Here we are reporting a case of Moyamoya disease of left petrous and cavernous part of internal carotid artery with contralateral distal anterior cerebral artery aneurysm in a 36-year-old male.

Key words: Digital subtraction angiogram (DSA), Moyamoya disease (MMD), Moyamoya syndrome (MMS), DACA aneurysm

Introduction

Moyamoya disease (MMD) is a chronic cerebrovascular disease of unknown etiology characterized by progressive occlusion of distal part of internal carotid artery (ICA) and circle of Willis and development of collateral vessels (moyamoya vessels) close to occlusion. [1] Unilateral moyamoya disease is known as Moyamoya syndrome (MMS). Progressive occlusion of ICA alters the flow dynamics in unobstructed vessels with compensatory dilatation of moyamoya vessels. This increase flow, result in rise of blood pressure and formation of aneurysm. [2] Reported incidence of intracranial aneurysm in MMD is 3.4-14.8 %. [3] Cerebral aneurysm with MMD is first reported in 1965. [4] Hemorrhagic event in MMD with associated aneurysm is believed to be due to rupture of aneurysm. [5]

Because of complex pathology and variable anatomy, even in the era of great advancement in micro neurosurgery and interventional neurology, clear guide lines for the management of aneurysm with MMD is lacking. We are reporting a case of MMD of left side petrous and cavernous segment ICA and aneurysm of right side distal anterior cerebral artery (DACA).

Case report

A 36-year-old male patient presented to our outpatient department with complaint of sudden onset severe headache, dizziness and nausea for 6 days with no past history of any systemic medical illness. Neurological examination at the time of admission was normal. Non-contrast CT shows intraventricular hemorrhage involving lateral, third and fourth ventricle. [Figures 1A and 1B] CT angiography revealed right side DACA aneurysm. Digital Subtraction angiogram was suggestive of MMD of left petrous and cavernous segment of ICA with aneurysm of pericallosal branch of DACA of size 4.6 mm with 2.18 mm neck. [Figures 2A and 2B] Patient underwent endovascular coiling embolization of the DACA aneurysm which was unsuccessful as catheterization of DACA was not feasible due to tortuosity of cervical segment of right ICA and right Anterior cerebral artery (ACA). Hence, microsurgical clipping was planned but the patient denied for clipping surgery. Patient was followed in outpatient department later for 6 months and then he lost the follow-up.

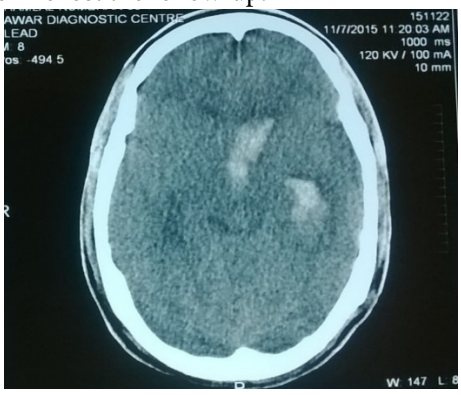

Figure 1A - CT head showing intraventricular haemorrhage

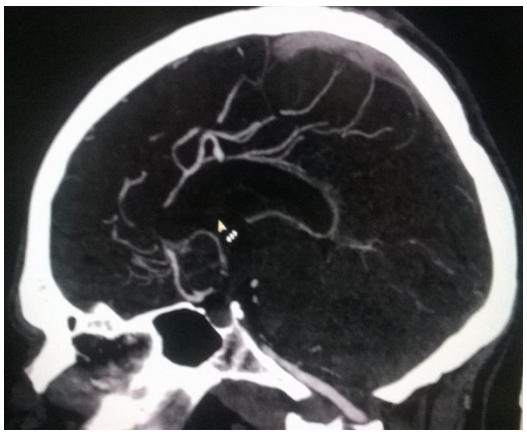

Figure 1B - CT angiography sagittal view showing DACA aneurysm

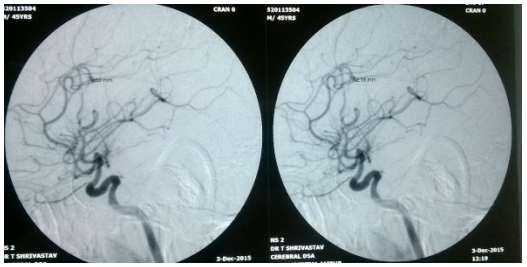

Figure 2A - DSA showing right side DACA aneurysm of size 4.6mm with 2.18 mm neck

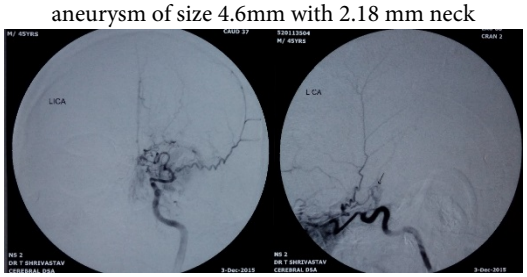

Figure 2B - DSA showing "puff of smoke" appearance characteristics of MMD of left Internal carotid artery (petrous and cavernous part)

Discussion

Simultaneous occurrence of MMD with intracranial aneurysms is reported by various authors from circle of Willis to moyamoya vessels, but posterior circulation aneurysms

are the most commonly reported aneurysm with MMD. [6] They are usually classified into two subtypes of MMD with aneurysm of major artery and MMD with aneurysm of peripheral artery. Major artery aneurysms are usually saccular aneurysm while peripheral artery aneurysms are pseudo aneurysm resulting from the rupture of fusiform or dissecting aneurysm arising from moyamoya vessels.

Yeon et al classified aneurysm with MMD into 3 categories, aneurysm at major arteries (circle of Willis), distal peripheral artery (anterior and posterior choroidal artery) and moyamoya vessels [7] MMD with associated meningeal artery aneurysm and anastomosis site aneurysm had also been reported in literature. [8] Kawaguchi et al reported 23% of anterior circulation aneurysm in his series of 111 patient of aneurysm with MMD. Treatment options for the anterior circulation aneurysm in MMD are greatly varied among the reported series and include either surgical clipping or endovascular embolization. [6] Over all outcomes of these aneurysm with associated MMD is not satisfactory with significant mortality and morbidity.

Direct surgical clipping as well as endovascular embolization of ruptured basilar tip aneurysm in MMD is associated with high risk of complication. Although Recent studies favours the endovascular embolization for basilar tip aneurysm in a patient with MMD, optimal strategy for the treatment is yet to define.

Treatment of anterior choroidal artery aneurysm in patient with MMD is based on the fact that whether patient will require revascularization procedure or not. In patient requiring revascularization procedure, craniotomy is preferred over endovascular treatment since it treats both the aneurysm and MMD simultaneously. Craniotomy and endovascular embolization, both are effective way to treat posterior choroidal artery aneurysm with associated MMD. Disappearance of posterior choroidal artery aneurysm following revascularization procedure as well as with conservative treatment has been reported in literature. [9]

Moyamoya vessel mainly signifies lenticulostriate artery (LSA) and thalamoperforator artery (TPA). There is various treatment option for the treatment of LSA aneurysm in MMD patients including surgical clipping, endovascular embolization and revascularization procedures. Each of these procedures has their own merits and demerits. Superficial temporal artery- Middle Cerebral artery (STA-MCA) anastomosis is an effective procedure for management of the ischemic lesion in MMD. Increase hemodynamic stress over anastomosis site makes it vulnerable for development of aneurysm. There are case reports on anastomosis site aneurysm which were treated with craniotomy and followed significant complications.

Iwama et al reported a case of MMD harbouring, three anterior circulation aneurysms located at A1 segment of ACA, Anterior communicating artery and DACA. Clipping was performed for all these aneurysms followed by STA-MCA bypass. [10] Postoperative outcome was satisfactory. This is the only case report of DACA aneurysm in patient with MMD available in

literature. However isolated DACA aneurysm in a patient with contra lateral MMD have never been reported in literature.

In nutshell, treatment of aneurysm with associated MMD depends on site and hemodynamic characteristic of aneurysm. Craniotomy is considered if revascularization procedure is required. Endovascular embolization is an alternate to craniotomy as it avoids the risk of craniotomy and risk of disruption of collateral vessels. Thus, revascularization surgery is one of the important factors to determine the outcome in aneurysm patient with associated MMD.

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