"Mickey Mouse head aneurysms" - kissing aneurysms of the distal anterior cerebral artery: a case report and literature review

Sachin Chemate^{1*}, K. Chandrasekar², Shankar Ganesh C. V.²,
Chetana Govindaraju¹, M. Balamurugan²

ABSTRACT

Background: When two or more adjacent intracranial aneurysms arises from same or different arteries with separate origins and partially adherent walls is referred as "Kissing aneurysm". These are rare aneurysms¹. Among different locations of kissing aneurysms reported in the literature, kissing aneurysms of bilateral distal anterior cerebral arteries (DACA) - two different aneurysms situated symmetrically on opposite DACA are very rare.

Case Description: 69 years old lady with multiple comorbidities was admitted with anterior inter-hemispheric bleed, Magnetic Resonance Angiography (MRA) and Digital Subtraction Angiography (DSA) showed ruptured bilateral DACA aneurysms in mirror position

to each other forming a Mickey Mouse head appearance. Patient was managed surgically by craniotomy and clipping of both aneurysms.

Conclusion: Kissing aneurysms of bilateral DACA aneurysms are very rare. DSA is essential to be useful for preoperative diagnosis and planning. Basic principle of securing the parent artery proximally and clipping the neck after meticulous dissection should be followed.

Key words: Kissing aneurysm, distal anterior cerebral artery aneurysm, Digital Subtraction Angiography.

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Cite This Article: Chemate, S., Chandrasekar, K., C.V.S.G., Govindaraju, C., Balamurugan, M. 2020. "Mickey Mouse head aneurysms" - kissing aneurysms of the distal anterior cerebral artery: a case report and literature review. *Indonesian Journal of Neurosurgery* 3(1): 33-37. DOI: 10.15562/ijn.v3i1.91

¹Resident, Department of Neurosurgery, Apollo Hospitals, Chennai

²Senior Consultant, Department of Neurosurgery, Apollo Hospitals, Chennai

*Corresponding Author:
Sachin Chemate; Resident,
Department of Neurosurgery,
Apollo Hospitals, Chennai
sachin.19chemate@gmail.com

INTRODUCTION:

'Kissing' aneurysms were initially defined and described by Jefferson¹ in 1978. When two or more adjacent Intracranial aneurysms arises from same or different arteries with separate origins and partially adherent walls is referred as "Kissing aneurysm".¹ They occur in <1% of all intracranial aneurysms.² The most common site is the internal carotid artery (ICA) followed by the anterior communicating artery (ACom), distal anterior cerebral artery (ACA), and fenestrated basilar artery (BA).²⁻⁸ Kissing aneurysms of bilateral distal anterior cerebral arteries (DACA) are rare and till date only 12 cases reports have been reported. We report one such case which was managed surgically by craniotomy and clipping of both aneurysms.

CASE REPORT

69 years old lady was admitted to emergency department with complaints of one episode of sudden loss of consciousness lasting for 5-10 minutes, followed by headache. She was known case of type II diabetes mellitus since 15 years and systemic hypertension since 10 years on

regular management and previous history of well differentiated squamous cell carcinoma of oesophagus diagnosed 5 years ago for which she underwent radiation and achieved remission. On examination, the patient was E4V4M6, vitals were stable, pupil was normal sized and reacting to light, and there were no focal neurological deficits. Due to previous history of carcinoma oesophagus and patient's history, cerebral metastasis was suspected and MRI brain was done but surprisingly it showed anterior interhemispheric bleed with corpus callosum hematoma (Figure 1a and 1b) and Magnetic resonance angiography (MRA) of the brain was suggestive of two adjacent aneurysm on bilateral distal anterior cerebral artery (ACA) at the junction of pericallosal and callosomarginal arteries (Figure 2a and d). Digital subtraction angiography (DSA) was done which showed two adjacent aneurysms over each DACA at the junction of pericallosal and callosomarginal arteries. Right side aneurysm was size 9.4 × 8.7 mm with a neck measuring 2.5 mm with a daughter lobule and superiorly projecting rupture site. Left-sided aneurysm measured size 7.6 × 5.5 mm with a neck of 2.5 mm. Both were directed antero-superiorly. Both A1 ACAs were of the same size with good

Figure 1a and 1b. T2W MRI brain showing anterior interhemispheric bleed

Figure 2a, 2b and 2c. MRA showing of two adjacent aneurysms over each DACA

Figure 3a, 3b, 3c and 3d: DSA showing of two adjacent aneurysms over each DACA

Figure 4. Navigation system showing aneurysm (green dot) pointed by white arrow.

crossflow. Focal areas of segmental spasm were noted in right pericallosal arteries (Figure 3a-d).

The patient underwent surgery; right frontal craniotomy was done. Anterior interhemispheric approach was utilized. With help of navigation (Figure 4), pericallosal and callosomarginal arteries of the left side was identified and traced proximally. Right DACA aneurysm arising from the junction of pericallosal and callosomarginal artery directing anterosuperiorly was visualized (Figure 5a and 5b). Temporary proximal clip was applied over right A2 and right DACA aneurysm was clipped. Left DACA aneurysm was identified, it was buried in the brain parenchyma with overlying hematoma and adhesions. Proximal control over right A2 achieved with temporary clip. While dissecting dome of aneurysm there was intraoperative rupture, which was managed with bipolar cautery and permanent clip applied at the neck of right DACA aneurysm. Intraoperative Indocyanine Green (ICG) dye showed patent proximal and distal DACA, both the aneurysms were completely secured and there was no residual neck (Figure 5c).

Minimal brain retraction was used. Postoperatively, patient was extubated immediately. Postoperatively patient had no new deficit. As our institutional protocol, patient was managed with triple-H therapy. CT brain was done on postoperative day 1, which showed no evidence of infarct or haemorrhage (Figure 6a and 6b). Patient was discharged on postoperative day 7 without any neurological deficit.

DISCUSSION:

Epidemiology

DACA aneurysms have been reported to have an average incidence of 4.4%.¹⁴ Bilateral DACA “mirror image aneurysms” are extremely rare⁹⁻²¹, with majority of reports from Japanese literature (Table 1).

Pathophysiology

Laitinen and Snellman²² postulated that a supreme anterior communicating artery, a bridging artery located at the bifurcation of A2 into the pericallosal and callosomarginal arteries and other embryological connections like Azygos ACA, and triple ACAs might represent an embryological remnant and lead to flow disturbance in anterior cerebral artery bifurcation causing bilateral, symmetrical aneurysms. However, no vascular anomaly was detected in our case. Yasargil and Carter^{23,24} treated 13 patients with DACA aneurysms with microsurgical techniques and found that in there were two cases where the aneurysmal sacs were connected. Based on this they suggested that, some

embryological variation, such as a supreme ACoA, may cause a flow disturbance leading to aneurysm formation. Jefferson¹ suggested hereditary basis for pathogenesis of bilateral DACA aneurysm but still remains unclear. Wanifuchi et al²⁵ described female dominance of bilateral DACA aneurysm but other multiple aneurysms also has higher frequency in females.

DIAGNOSIS

CT angiography, MRA, and DSA are most useful

for the diagnosis of bilateral DACA aneurysms. Although MRA is very sensitive and specific for distal aneurysm and 3D-reconstructions are also available in this imaging, however, DSA provides complete study of intracranial vessels in real time. Vasospasm can also be diagnosed during angiogram in early phases. Other distal Incidental aneurysms can be seen in angiograms. It also gives very critical information regarding aneurysm location, shape, size of neck, branch vessel, and cross circulation; which helps surgeon to decide whether to go for clipping or coiling.

Figure 5.a and 5.b. intraoperative images showing mirror DACA aneurysms

Figure 6a and 6b. CT brain showing clips applied on both DACA aneurysm

Management

The most definite management at the bilateral DACA aneurysm is clipping the aneurysm. Bilateral DACA aneurysm has tendency to rupture so it presents with subarachnoid haemorrhage and anterior interhemispheric hematoma. Incidence of rebleed is also very high in DACA aneurysm. Through anterior interhemispheric approach we can not only achieve a proximal control on A2 and secure the aneurysm but also it provides an access evacuation of for hematoma and mass effect and thorough washing of cranial cavity with normal saline during surgery also removes blood by products which decreases the intensity and duration of vasospasm. Aneurysm occlusion rate is very high with clipping and rebleeding or recurrence is very rare after clipping. Challenges of craniotomy and clipping are narrow working corridor, edematous brain, adhesions and buried dome of aneurysm in the surrounding brain parenchyma. Dome of the aneurysms is always directed towards the surgeon and proximal A2 is inferior to aneurysm when visualised from working corridor, hence proximal

Table 1.

No.	Year	Author	Case reports
1	1988	Megele et al ⁹	A case of symmetrical pericallosal aneurysms with recurrent haemorrhage
2	1989	Nijima et al ¹⁰	Bilateral pericallosal artery aneurysms in a mirror position
3	1995	Mori et al ¹¹	Kissing aneurysms of DACA demonstrated by magnetic resonance angiography
4	2001	Moon et al ¹²	Kissing aneurysms of DACA distal anterior cerebral arteries
5	2002	Sousa et al ¹³	Mirror image DACA aneurysms. A case report of two patients with review of literature
6	2003	Kawamata et al ¹⁴	Bilateral DACA aneurysms associated with supreme anterior cerebral artery: case report
7	2006	Ahn et al ¹⁵	Kissing aneurysms of DACA
8	2006	Dinc et al ¹⁶	DACA mirror aneurysms and middle cerebral artery aneurysms
9	2010	Meysam Alimohammadi et al ¹⁷	Bilateral "Kissing" Aneurysms of the Distal Pericallosal Arteries
10	2011	Choi CY et al ¹⁸	Kissing aneurysms of the distal anterior cerebral artery
11	2013	E. Enesi et al ¹⁹	Mirror Image Distal Anterior Cerebral Artery Aneurysms Treated with Coil Embolization
12	2018	Chuan-Yi Fu et al ²⁰	Kissing aneurysms of the distal anterior cerebral artery.
13	2018	Singh SK ²¹	Mirror image of bilateral DACA aneurysm with its successful surgical management.

control is challenge and chance of intraoperative rupture is high. Only one case report is available in literature about the experience of coiling.

Our patient was taken up for clipping after angiogram. During the operative procedure, retraction was applied first on the right medial frontal lobe. Parent artery and dome of left aneurysm was identified. Dome was followed inferiorly up to the neck and the clip was applied on proximal A2. After securing left-sided aneurysm completely, right-sided A2 proximal clip was applied. Dome and neck of the right DACA aneurysm was then dissected and clip applied. However while dissecting the aneurysm aneurysm ruptured in spite of temporary clip on proximal A2. Recurrent use of temporary clips loses there occlusion force resulting in only partial occlusion. Intraoperative rupture was managed with bipolar cautery over dome and multiple clipping over the bleb. After securing the bleb rupture, final clip was applied at the neck.

CONCLUSION

DSA is essential for diagnosis of multiple intracranial aneurysm. Mirror images of bilateral DACA aneurysms are very rare. They are specific type of aneurysms with probably derived from embryological rearmament. Basic surgical strategy of securing the parent artery and clipping the neck after meticulous dissection of each aneurysm separately is key for successful clipping of aneurysm, as chance of intraoperative rupture is very high.

CONFLICT OF INTEREST

There are no conflicts of interest.

FINANCIAL SUPPORT AND SPONSORSHIP

There is no funding available for this case report.

AUTHORS CONTRIBUTIONS

All authors equally contributed immensely.

ABBREVIATION

DACA - Distal Anterior Cerebral Arteries
MRA - Magnetic Resonance Angiography
DSA - Digital Subtraction Angiography
ACoM - Anterior Communicating Artery
ACA - Anterior Cerebral Artery
BA - Basilar Artery
Fig - Figure
ICG - Intraoperative Indocyanine Green
CT - Computed Tomography

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