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Case Report

Ruptured aneurysm associated with a twig-like middle cerebral artery: An illustrative case report

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ABSTRACT

Background: Anomalies of the middle cerebral artery (MCA) are rare; among the different types of anomalies, the aplastic or twig-like (Ap/T) MCA is extremely rare and has been reported under various names, including aplastic, unfused, or rete type anomaly. The occurrence of a brain aneurysm associated with this anatomic variant is an even rare event, and probably their development and rupture are related to hemodynamic stress of the tinny wall of vessels forming the network.

Case Description: We present a 43-year-old male patient with an explosive and persistent right orbitofrontal headache. A computed tomography showed a right frontobasal hematoma with intraventricular disruption. Magnetic resonance angiography showed a right MCA aneurysm and what seems to be a MCA trunk stenosis. Cerebral digital subtraction angiography demonstrated a plexiform arterial network and one aneurysm arising from the network. The patient was successfully treated by surgical clipping to evacuate the hematoma and to prevent further intracranial hemorrhages.

Conclusion: The Ap/T-MCA may be associated with hemodynamic stress with a significant effect through the tinny wall of the vessels causing hemorrhage or leading to the formation and rupture of cerebral aneurysms. Based on a correct diagnosis of the anomaly, treatment can be completed successfully through different standard methods.

Keywords: Aneurysm, Clipping surgery, MCA anomaly, Middle cerebral artery, Twiglike MCA

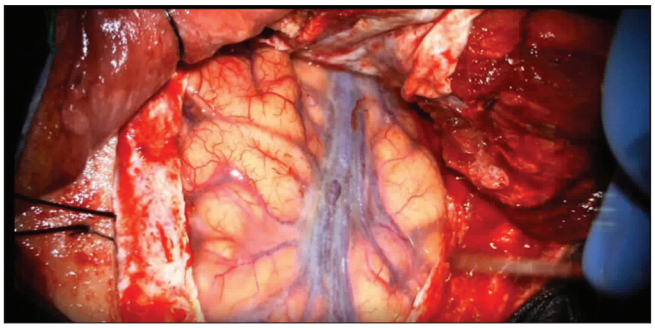
INTRODUCTION

Aplastic or twig-like middle cerebral artery (Ap/T-MCA), also called "unfused MCA," "rete MCA" or "anomalous collateral artery," is a rare anatomic anomaly with a very low prevalence among population (0.11–0.88%),[4] which results in the formation of a plexiform arterial network replacing the proximal M1 segment, resembling sometimes the collateral network present in patients with Moyamoya disease (MMD), with the exception that the internal carotid artery stenosis is absent.[27] On magnetic resonance imaging (MRI) and magnetic resonance angiography (MRA), this anomaly may be confused with a stenotic segment of the MCA. Ap/T-MCA may cause ischemic or hemorrhagic events. In the last case, these may occur spontaneously or because the rupture of an associated cerebral aneurysm. However, this association is rather a rare event. After the first report of Yasargil *et al*. in 1976, there exist only a few reports in the literature about this association and the best way of treatment.[35]

CASE DESCRIPTION

This 43-year-old male with a history of systemic arterial hypertension was admitted to the hospital because a sudden onset of severe headache of 7 days evolution accompanied by nausea, vomiting, and a right-side hemiparesis.

Computed tomography (CT) revealed a frontobasal hematoma with ventricular disruption [Figure 1a]. A CT angiography (CT-Angio) and a cerebral digital subtraction angiography (DSA) were completed, revealing an abnormal network of plexiform vessels with a "twig-like" appearance and the presence of a saccular aneurysm of 9 mm of diameter at the M1 segment of the right MCA [Figures 1b and c].



Video 1: Aplastic or twig-like middle cerebral artery. An illustrated case report.

The patient was operated on 3 weeks after ictus, through a right side pterional approach. After opening the Sylvian fissure, the vessels network was visualized, formed by numerous tinny and fragile vessels, with the aneurysm located in the middle of this network with old clots and hemosiderin surrounding the lesion [Figure 1d]. During the dissection of the vessels surrounding the aneurysm, the slight movements of the branches caused them to break easily due to the fragility of their walls, which caused profuse bleeding, confusing it with a probable aneurysmal rupture; however, the bleeding points stopped easily by the pressure with surgical patties for 1–2 min.

A straight 7 mm Yasargil type clip was used for neck clipping. We verified total occlusion using intraoperative fluorescein videoangiography and intraoperative Doppler. The postoperative course was uneventful and the patient showed a total recovery of the initial preoperative neurologic deficit in the next days. Postoperative cerebral DSA and CT-

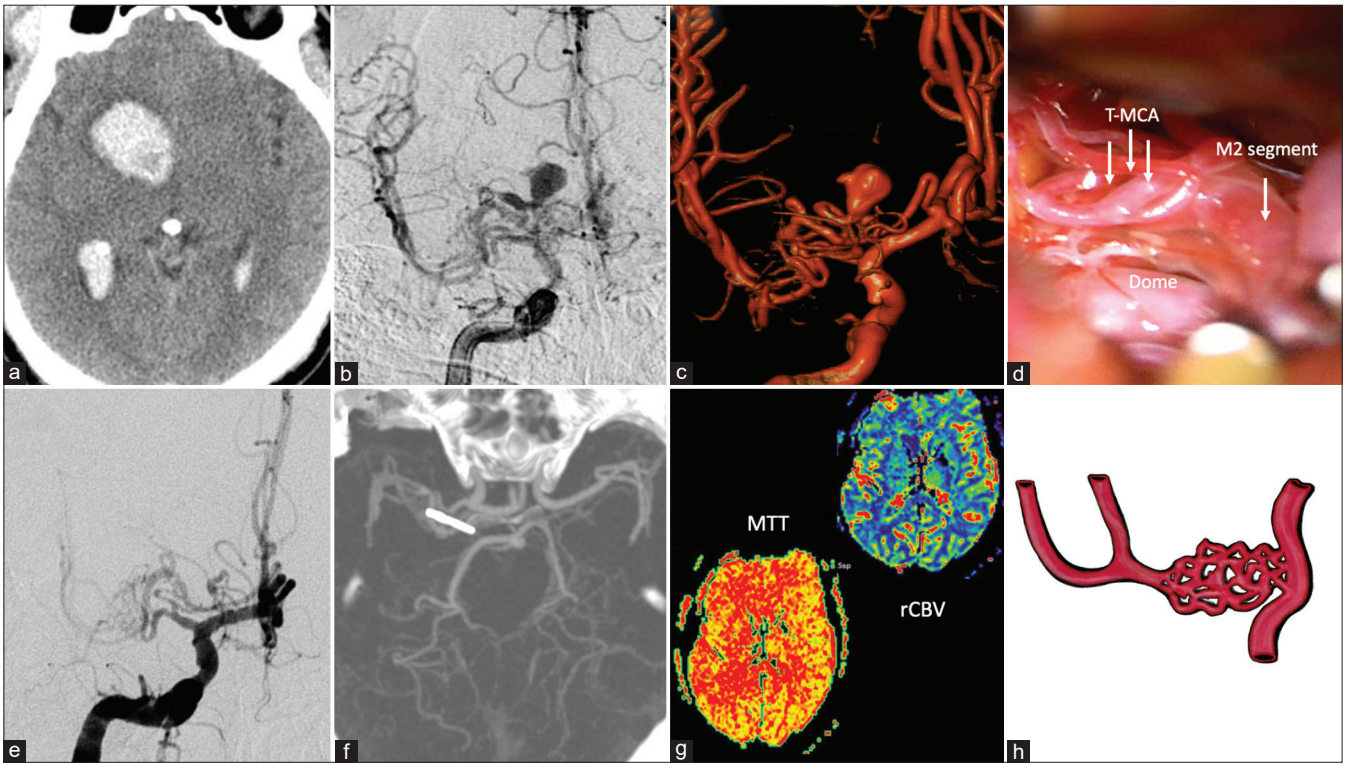


Figure 1: Illustrative case (a) A right frontobasal hematoma with ventricular extension is seen on noncontrast Computed tomography (CT) scan. (b and c) Cerebral DSA with rotational 3-D show the presence of a saccular aneurysm in the middle of the vascular network. (d) Intraoperative view. After evacuating the hematoma, the vascular network is exposed (T-MCA). Under the superficial vessels of the anomaly, the dome of the aneurysm is just visible. Distal M2 segment seems normal. (e) Postoperative DSA. The aneurysm is not visible. (f) CT-angio displays the clip position and the complete exclusion of the aneurysm. (g) Perfusion MRI showing the mean transit time (MTT) and regional cerebral blood volume (rCBV) images without ischemic areas (h) Illustration of the Ap/T MCA anomaly depicting the plexiform arterial network replacing the proximal M1 segment.

Table 1: Published cases of Ap/T-MCA associated with brain aneurysm.

Case No.	Authors	Year	Age, Sex (years)	Clinical presentation	Aneurysm			Treatment	Follow-up
					No.	Location	Site		
1	Yasargil *et al.*[35]	1976	38/M	SAH	1	ACoA	Rt	Clipping	N/A
2	Tanaka *et al.*[30]	1980	40/M	SAH, IVH	1	PCA	Lt	Clipping	6 Mo
3	Fukawa *et al*[6]	1982	53/F	SAH	1	Pcho	Lt	Not available	N/A
4	Matsuda *et al.*[16]	1983	51/F	IVH	2	PLcho ICA	Lt Rt	Trapping, EMS, Conservative	2 Mo
5	Nakazawa *et al.*[17]	1985	40/M	SAH, ICH	1	ACA	Lt	Clipping	3 Mo
6	Grabel *et al.*[9]	1989	60/M	ICH	1	M_1	Lt	Conservative	3 weeks
7	Kageji *et al.*[12]	1992	23/M	SAH	1	MCA	Lt	Clipping	N/A
8	Han *et al.*[10]	1994	34/F	SAH	1	ACA	Lt	Clipping	10 days
9	Inoue *et al.*[11]	1994	59/M	SAH	1	ACA	Rt	Clipping	3 Mo
10	Amagasaki *et al.*[2]	1998	64/M	Incidental	1	A_2	Lt	Clipping	N/A
11	Seki *et al.*[24]	2001	63/F	ICH, SAH	2	A_1 P_2	Lt Lt	Clipping, Trapping	23 Mo
12	Park *et al.*[20]	2004	74/F	ICH, SAH	2	M_1 M_1	Rt Rt	Clipping, Clipping	N/A
13	Kim *et al.*[13]	2005	64/F	SAH	2	A_1 ACoA	Lt Lt	Clipping, Clipping	Died (12 days)
14	Liu *et al.*[15]	2005	44/M	ICH, SAH	1	M_1	Lt	Clipping	N/A
15			67/F	ICH, SAH	1	M_1	Lt	Clipping	N/A
16	Cekirge *et al.*[3]	2005	32/M	SAH, IVH	1	A_1	Lt	Coiling	18 Mo
17	Sakai *et al.*[23]	2005	65/F	ICH	1	M_1	Rt	Clipping & bypass	N/A
18	Narisawa *et al.*[18]	2009	83/M	ICH	1	MCA	Lt	Trapping	N/A
19	Rodríguez-Hernández *et al.*[21]	2011	52/M	ICH	1	M_1	Rt	Trapping	3 Mo
20	Seo *et al.*[26]	2012	49/F	ICH	1	SHA	Rt	Burr hole, coiling, EDAS	N/A
21			58/M	Incidental	2	SHA SHA	Lt Rt	Conservative, Coiling	N/A
22			73/M	SAH	1	MCA	Rt	Clipping	N/A
23			58/F	Acute infarction	1	ICA	Lt	Conservative	N/A
24			73/M	SAH	1	ICA	Rt	Conservative	N/A
25	Shin *et al.*[27]	2014	42/M	SAH, ICH, IVH	2	M_1(1) M_1(2)	Lt Lt	Trapping, Clipping	26 Mo
26			49/F	SAH, ICH, IVH	1	M_1	Lt	Clipping	26 Mo
27			46/F	SAH, ICH, IVH	2	M_1 A_1	Lt Lt	Gluing, Clipping	26 Mo
28			26/F	SAH, IVH	1	PLcho	Rt	Observation	26 Mo
29	Akkan *et al.*[1]	2015	54/M	Incidental	1	ICA	Lt	Conservative	6 Mo
30	Uchiyama *et al.*[31]	2017	77/F	ICH, IVH	1	MCA	Lt	Conservative	35 Mo
31	Lang *et al.*[14]	2017	61/M	ICH, IVH	1	M_1	Rt	Trapping	2 yrs
32			53/F	SAH	1	MCA	Lt	Clipping	2 yrs
33	Seno *et al.*[25]	2017	49/F	SAH, IVH	1	AChA	Rt	Clipping & bypass	2 yrs
34	Fukuda *et al.*[7]	2018	60/F	SAH	1	M_2	Lt	Coiling	2 yrs
35	Fukuyama *et al.*[8]	2020	53/F	SAH	1	M_1	Lt	Clipping	2 yrs
36	Yamada *et al.*[34]	2020	68/F	CI, ICH	1	MCA	Lt	Bypass	2 yrs
37	Viso *et al.*[32]	2021	59/M	SAH, IVH	1	AcoA	Lt	Coiling	3 yrs
38			48/F	SAH, IVH	1	AcoA	Rt	Coiling	3 yrs
39			48/F	ICH	1	AcoA	Rt	Coiling	3 yrs
40			42/F	ICH	1	PcomA	Lt	Coiling	3 yrs
41			64/F	IVH	1	ACA	Rt	Clipping	3 yrs
42	Takarada *et al.*[29]	2021	46/F	IVH	1	MCA	Lt	Clipping & bypass	6 Mo
43	Present case	2022	43/M	SAH	1	MCA	Rt	Clipping	3 Mo

EDAS: Encephaloduroarteriosynangiosis, EMS: Encephalomyosynangiosis, PCA: Posterior cerebral artery, Pcho: Posterior choroidal artery, PLcho: Posterolateral choroidal artery, SHA: superior hypophyseal artery, AcoA: Anterior communicating artery, ACA: Anterior cerebral artery, PcomA: Posterior communicating artery, MCA: Middle cerebral artery, ICA: Internal carotid artery, ICH: Intracerebral hemorrhage, SAH: Subarachnoid hemorrhage, IVH: Intraventricular hemorrhage, Rt: Right, Lt: Left

angio showed the disappearance of the aneurysm without associated low-density areas [Figures 1e and f] and the perfusion MRI did not show any ischemic area [Figure 1g]. Patient will be maintained in a long-term follow-up to evaluate the possibility of new aneurysms or ischemic symptoms through CT-angio or MRA [Video 1].

DISCUSSION

Ap/T-MCA is a rare vascular anomaly. It has been considered that embryological interruption of the MCA trunk genesis is the cause, being replaced by the formation of a plexiform arterial anomaly [Figure 1h]. This anomaly has been related with ischemic or hemorrhagic events; however, there is a very low information about the association with cerebral aneurysms as the cause of hemorrhagic episodes.[28,31]

Due to the difficulty of diagnosing this anomaly on noninvasive imaging modalities such as CT and MR angiography, it is often misdiagnosed as arteriovenous malformation or pseudo-occlusion of the MCA. In patients that underwent a DSA, the Ap/T-MCA was present 0.11% and 1.17% of them.[15,26]

Akkan *et al*. described three types of "Ap/T-MCA" presentations, where the entire MCA is composed of a vascular network; in another presentation, the vascular network terminates at the bifurcation. In the last case, the vascular plexiform network ends before the beginning of the bifurcation.[1] Analyzing the DSA of our patient, we conclude that the Ap/T-MCA ended before the beginning of the bifurcation.

The twig-like appearance of the vessels may be confused with the MMD; however, there is no evidence of a progressive internal carotid artery occlusion leading to the formation of the Moyamoya vessels, as occurs in MMD.[19]

At present, there are 42 cases in the international literature of aneurysms associated with the Ap/T-MCA anomaly, of which 26 presented as SAH,[3,4,6-8,10-15,17,20,24-28,30,32,35] 27 associated with intracerebral[9,14,15,17,18,20,21,23,24,26,27,31,32,24] or intraventricular hemorrhage,[3,14,16,25,27,29-32,35] three incidental,[2,26,1] and one with a cerebral infarction[26] [Table 1]. The aneurysm formation is considered to be caused by hemodynamic stress on the twig-like tinny vessels leading to rupture and hemorrhage. In two of these cases, pathologic studies reported an internal elastic lamina disruption at the neck and wall disruption at the level of the dome of the aneurysm as the cause of rupture with a surrounding hematoma, similar as occurs in other brain aneurysms.[5,22,27,33]

In this case, our patient had a Fisher Grade IV subarachnoid hemorrhage. During surgery, after evacuating the surrounding hematoma, we found many fragile vessels of the network around the aneurysm that bleed easily. Compression of the bleeding points with surgical patties quickly stopped the hemorrhage without the need of cauterization with the bipolar forceps, assuming the thrombotic mechanism of the vessel.

Ap/T-MCA can present with hemorrhagic stroke in 27–40% of the cases, of which up to 26.6–46% were associated with aneurysm rupture. On the other hand, ischemic stroke can be present in 33–46% of cases.[23] Although this vascular network is functional, the patient should be warned about the nature of the anomaly due to the risk of rupture or ischemia or development of cerebral aneurysms. The utility of antiplatelet drugs or reduction of the hemodynamic stress through a revascularization procedure (like the STA-MCA bypass) is still a matter of concern due to the scarcity of information about the long-term course of these patients.[27]

Endovascular coil embolization and surgical clipping are procedures that have been associated with success in managing ruptured aneurysms. However, in the presence of a hematoma causing a mass effect, as occurred in this case, the surgical approach is mandatory. Endovascular coiling has been used mainly in aneurysms located lateral to the twig-like anomaly. In cases associated with ischemic events, cerebral revascularization surgery may be used whenever a perfusion study was done showing hypoperfusion in the MCA territory of the Ap/T-MCA. In our case, this procedure was unnecessary since there were not ischemic events or neurological deficits.[20,11] The use of intraoperative ancillary methods such as Fluorescein videoangiography or intraoperative Doppler may increase the confidence of the surgical procedure to avoid postoperative ischemic complications.[33]

CONCLUSION

The Ap/T-MCA is a rare anomaly. There is a lack of knowledge and information about it due to the scarcity of reports in the literature. However, when present, it should be diagnosed correctly to avoid unnecessary therapeutic considerations in patients with acute symptomatology. DSA remains as the gold standard for diagnoses. The occurrence of cerebral aneurysms associated with this anomaly requires additional considerations. In case of unruptured aneurysms, an endovascular or surgical procedure has been reported as effective methods, but in ruptured cases associated with brain hematomas, surgical treatment is mandatory. This case gives rise to a complete opening in both diagnosis and treatment of aneurysms in variants of the MCA as the twig-like type. More detailed clinical studies with a large population are required to determine the optimal treatment of Ap/T-MCA associated with aneurysms or ischemic events.

Declaration of patient consent

Patient's consent not required as patient's identity is not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

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