



Rete Middle Cerebral Artery Aneurysm: A Case Report and Systematic Review

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Asian J Neurosurg 2023;18:790–795.

Abstract

Keywords

- ▶ MCA anomaly
- ▶ rete MCA
- ▶ twig-like MCA
- ▶ anomalous collateral artery
- ▶ aneurysm

Background Middle cerebral artery (MCA) anomalies are a rare finding and may be associated with vascular changes, such as intracranial aneurysms. Among them, the rete MCA aneurysm is very rare, with only 22 cases reported to date.

Case Description A 50-year-old woman presented with subarachnoid, intraventricular, and intracerebral hemorrhage secondary to a ruptured aneurysm of rete MCA from an anomalous collateral artery of the anterior cerebral artery, treated successfully by microsurgical clipping. She presented a good recovery after a 2-year follow-up.

Conclusion A systematic review of rete MCA aneurysms is presented, comparing aneurysms originating from twig-like MCA, with 16 reports, and twig-like networks of an anomalous collateral artery, with 6 reports including ours. Several factors influence the treatment decision-making, though microsurgical clipping is the main procedure. A wider use of coiling is requested for a better comparison of the treatment approaches.

Introduction

Middle cerebral artery (MCA) anomalies are a rare finding. Among them, a rete MCA is extremely rare and has not been frequently discussed. This is a weblike anomaly of the MCA, architected as a tortuous, large single branch of plexiform vessels, and may be associated with intracranial aneurysms,^{1,2} with only 22 case reports of aneurysms located in a rete MCA in the literature. The pathogenic mechanism of rete MCA remains unclear. The main presentation of rete MCA is associated with the formation of twig-like networks in the MCA main trunk (T-MCA), with 16 reports to date.

Another singular occurrence is a rete MCA originating from an anomalous collateral artery, that is, twig-like networks of an anomalous collateral artery (T-NACA),² with only six reports including ours.

The purpose of this study is to present a case of ruptured aneurysm associated with rete MCA from an anomalous collateral artery of the anterior cerebral artery (ACA), to discuss the clinical, imaging, and surgical characteristics, and to review the literature on the subject of rete MCA aneurysms. A detailed, updated discussion on aneurysms of rete MCA from an ACA anomalous collateral artery is also presented.

article published online
November 7, 2023

DOI <https://doi.org/10.1055/s-0043-1775732>.
ISSN 2248-9614.

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Case Presentation

A 50-year-old woman was admitted to the emergency department with acute onset of severe headache associated with multiple vomiting episodes and no history of trauma. The neurological examination revealed disorientation, aphasia, and right hemiplegia (Hunt and Hess score 4). She had been receiving aspirin therapy for 10 years due to systemic arterial hypertension, without using another drug. There was no personal or family history of cerebrovascular disease.

Her computed tomography revealed subarachnoid hemorrhage, intraventricular hemorrhage (IVH), and left frontal intracerebral hemorrhage (ICH), without hydrocephalus (modified Fisher scale score 4). Digital subtraction angiography revealed a twig-like network, a rete MCA from an ACA anomalous collateral artery, and a small saccular aneurysm (2.3 mm) (►Fig. 1). She had no previous angiography for comparison.

By a pterional approach, two left MCAs were identified intraoperatively: an atretic M1 and an ACA anomalous collateral artery (►Fig. 2). Of these, the ACA anomalous collateral artery presented the largest caliber and an extensive network of plexiform branches (rete MCA anomaly). The frontal hematoma was evacuated, and the plexiform vessels were dissected, in which a small aneurysm was identified. The clipping was performed with a 7-mm straight clip. There were no intraoperative complications.

During the postoperative period, she evolved with persistence of aphasia and right hemiplegia. She left the hospital one week later, with a Glasgow outcome scale (GOS) score 3. During a period of 3 months, she evolved well, with partial resolution of the symptoms (GOS score 4). After a 2-year follow-up, she presented a good recovery, with slight dysphasia (GOS score 5). Her 2-year control angiography demonstrated an intact clipping instrumentation.

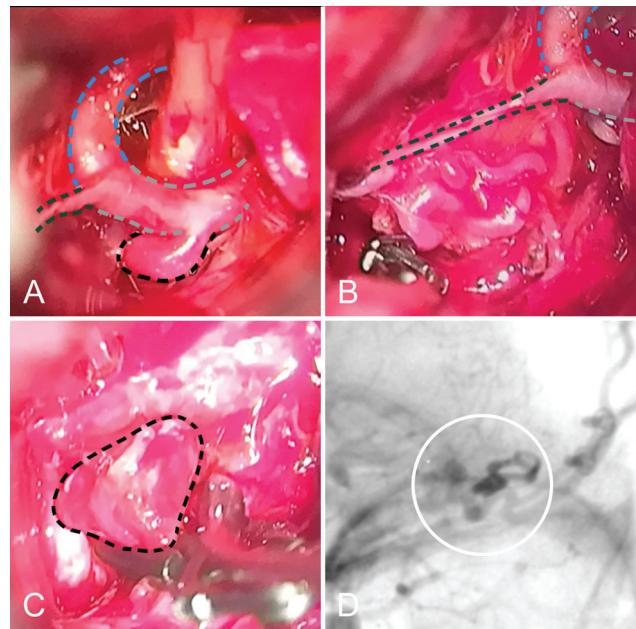


Fig. 2 Intraoperative images. (A) Visualization of internal carotid artery (blue), atretic middle cerebral artery (MCA) (green), anterior cerebral artery (gray), and rete MCA (black). (B) Twig-like, plexiform branches of the rete MCA, with diffuse subarachnoid hemorrhage, and atretic MCA (green). (C) After evacuation of the left frontal intracerebral hemorrhage and dissection of the plexiform vessels, a small aneurysm was identified (black) and clipped with a 7-mm straight clip. (D) Angiography demonstrating a twig-like network (rete MCA), without aneurysm (clipped) (white circle), after a 2-year follow-up.

Literature Review

Systematic Review Method

Following the standard PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) guidelines, we performed a systematic review of the available literature reporting cases of aneurysms located in a rete MCA. PubMed,

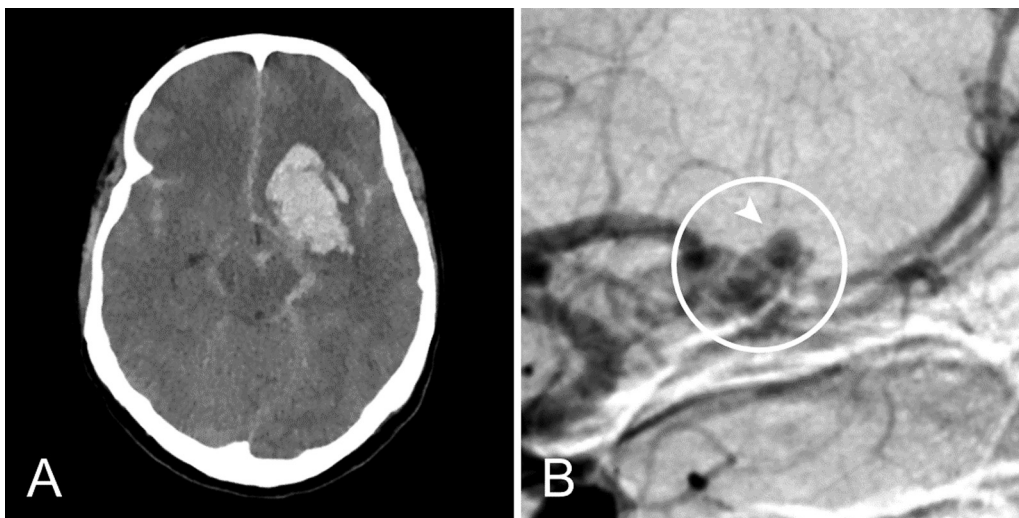


Fig. 1 Preoperative images. (A) Computed tomography scan demonstrating diffuse subarachnoid hemorrhage with intracerebral hemorrhage in the left frontal lobe, without hydrocephalus or intraventricular hemorrhage, scored as modified Fisher 4. (B) Angiography demonstrating a twig-like network (rete middle cerebral artery) (white circle) and a small (2.3 mm) saccular aneurysm (white arrowhead).

Scopus, Virtual Health Library, and ScienceDirect were queried from inception until June 2023. The search strategy (including title and abstract) was carried out as follows: (“rete” OR “twig-like” OR aplastic OR “anomalous collateral” OR “moyamoya”) AND (“MCA” OR “middle cerebral artery” OR “lenticulostriate”) AND “aneurysm”. We excluded cases of aneurysms not located in the rete MCA; patients with aneurysms located in the contralateral MCA of the rete anomaly; unavailable text; articles not published in English language; and abstracts that were not followed by a peer-reviewed publication. Two authors independently extracted data (JC-N and GM) following predefined search criteria and quality assessment methods. After applying the exclusion criteria, four reviewers (JC-N, GM, AB, and GS) analyzed the remaining articles for quality assessment. Disagreements between these authors were resolved by consensus among all authors until agreement was achieved.

Review Results

A total of 2,703 relevant publications were identified. 511 duplicates were removed, remaining 2,192 articles. Subse-

quently, 2,170 studies were removed after reading the title and abstract. Then, 22 articles were selected for full-text reading. In sequence, 8 articles were excluded after full-text reading. One article was manually included by cross-referencing.³ Ultimately, 15 articles were included, with 21 cases presenting similar characteristics. ►**Fig. 3** details the search strategy result.

Including ours, only 22 cases with a good description have been reported in the English literature, with a total of 28 rete MCA aneurysms (►**Table 1**).¹⁻¹⁵ ►**Table 1** presents a summary of rete MCA aneurysms, enabling a comparison between T-MCA and T-NACA aneurysms. The average age of patients is 56.1 years, with no gender prevalence (11/22, 50%). Most patients were Asians (16/22, 72.7%). All patients presented bleeding at admission (i.e., no incidental finding). The rete MCA had a slight prevalence by the right side (12/22, 54.5%). All aneurysms were small (28/28), and the majority presented a saccular morphology (27/28, 96.4%). The most common location was the M1 segment (26/28, 92.8%), in which almost half the aneurysms were located in the lenticulostriate arteries (10/26, 38.4%). When compared to

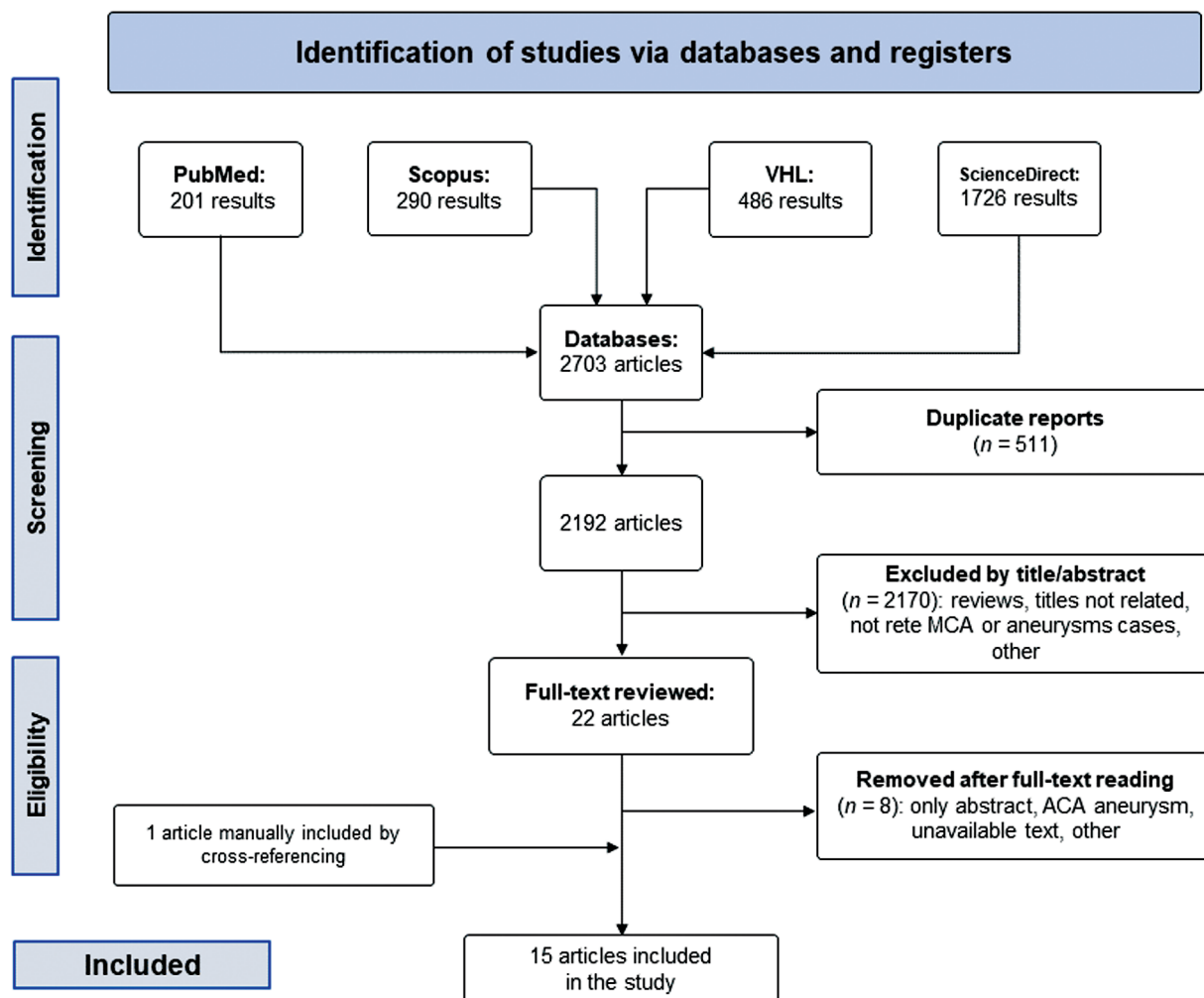


Fig. 3 Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flow diagram resuming the literature review process. ACA, anterior cerebral artery; MCA, middle cerebral artery; VHL, Virtual Health Library.

Table 1 Comparison of clinical, surgical, and outcome factors of rete MCA aneurysms

Variable	“T-MCA” aneurysms	“T-NACA” aneurysms	Overall rete MCA aneurysms
No. of cases	16	6	22
Average age	57 years (42–77)	53 years (38–74)	56.1 years (38–77)
Sex	6F:10M	5F:1M	11F:11M
Presentation	SAH (8/16, 50%), ICH (12/16, 75%), IVH (5/16, 31.2%)	SAH (4/6, 66.6%), ICH (6/6, 100%), IVH (4/6, 66.6%)	SAH (12/22, 54.5%), ICH (18/22, 81.8%), IVH (9/22, 40.9%)
Side	10R:6L	2R:4L	12R:10L
Treatment	Clipping (9/16, 56.2%), PVS (3/16, 18.7%), conservative (3/16, 18.7%), resection (1/16, 6.2%)	Clipping (4/6, 66.6%), PVS (1/6, 16.6%), resection (1/6, 16.6%)	Clipping (13/22, 59%), PVS (4/22, 18.1%), conservative (3/22, 13.6%), resection (2/22, 9%)
Outcome	Good recovery (11/12, 91.6%), bad recovery (1/12, 8.3%)	Good recovery (4/5, 80%), bad recovery (1/5, 20%)	Good recovery (15/17, 88.2%), bad recovery (2/17, 11.7%)

Abbreviations: ICH, intracerebral hemorrhage; IVH, intraventricular hemorrhage; MCA, middle cerebral artery; PVS, parent vessel sacrifice (by trapping, proximal occlusion, or distal occlusion); SAH, subarachnoid hemorrhage; T-MCA, twig-like MCA; T-NACA, twig-like networks of an anomalous collateral artery.

other MCA anomalies, ruptured rete MCA aneurysms presented a significantly higher rate of ICH (88.9 vs. 8–20%) and IVH (44.4 vs. 0–5%),¹⁶ due to a more distal location, with frequent implication of lenticulostriate arteries.

As for accompanying intracranial anomalies in rete MCA aneurysms, most patients (15/22, 68.1%) presented other reported concurrent cerebrovascular anomalies: steno-occlusive disease of the MCA or other vessels (13/22, 59.1%), anomalous collateral artery (6/22, 27.2%), and multiple aneurysms (6/22, 27.2%). Three patients (3/22, 13.6%) presented two aneurysms in the rete MCA,^{1,2,9} while two patients (2/22, 9%) presented another aneurysm in a different vessel,^{1,2} and one patient (1/22, 4.5%) presented with

four aneurysms in the rete MCA.¹⁴ To date, only six cases comprehended aneurysms of rete MCA from an anomalous collateral artery, as depicted in **Table 2**, of which five (4/6, 66.6%) extended from the A1 segment, while two (2/6, 33.3%) from the A2.^{2,7,9}

The main approach for rete MCA aneurysms is microsurgical clipping (21/28, 75%). Reported alternative treatments parent vessel sacrifice, conservative, and aneurysm resection. The parent vessel sacrifice (by trapping, proximal occlusion, or distal occlusion) was decided for four patients, due to initial procedure failure or operator choice.^{2,5,7,10} The conservative treatment was decided for three patients, due to aneurysm disappearance or initial procedure failure.^{2,4,6}

Table 2 Characteristics of aneurysms associated with rete MCA from anomalous collateral artery (i.e., T-NACA)

Case	Year	Authors [reference] (country)	Age/sex	Presentation	Aneurysm (no, location, site)	Accompanying intracranial anomalies	Treatment	Complication	Outcome
1	2004	Park et al ⁹ (South Korea)	74 years/F	SAH, ICH	02, M1, right	ICA bifurcation atresia, A2-M1 anomalous CA, T-NACA	Neck clipping (x2)	No	NA
2	2014	Shin et al ² (South Korea)	49 years/F	SAH, ICH, IVH	01, M1, left	M1 atresia, A2-M1 anomalous collateral artery, T-NACA	Proximal clipping	No	Good recovery
3	2014	Shin et al ² (South Korea)	46 years/F	SAH, ICH, IVH	01, LSA, left	M1 atresia, A1-M1 anomalous collateral artery with unruptured aneurysm, T-NACA	Gluing, neck clipping	Yes (gluing failure)	Bad recovery
4	2017	Lang et al ⁷ (USA)	61 years/M	ICH, IVH	01, LSA, right	M1 atresia, A1-M1 anomalous collateral artery, T-NACA	Proximal PVS	Yes (shunt infection)	Good recovery
5	2022	Watanabe et al ¹⁵ (Japan)	38 years/M	ICH	01, M1, right	M1 atresia, A1-M1 anomalous collateral artery, T-NACA	Resection	No	Good recovery
6	2023	Present case (Brazil)	50 years/F	SAH, ICH, IVH	01, M1, left	M1 atresia, A1-M1 anomalous collateral artery, T-NACA	Neck clipping	No	Good recovery

Abbreviations: ICH, intracerebral hemorrhage; IVH, intraventricular hemorrhage; LSA, lenticulostriate arteries; NA, not available; PVS, parent vessel sacrifice; SAH, subarachnoid hemorrhage; T-NACA, twig-like networks of an anomalous collateral artery.

Note: The anomalous collateral artery extended from the anterior cerebral artery in all cases.

One patient underwent aneurysm resection without vessel sacrifice, due to rebleeding after a conservative approach.⁵

To date, five procedure-related complications of rete MCA aneurysms have been reported (5/26, 19.2%), including procedure failure, rebleeding, and shunt infection.^{2,5,7,10} The prognosis of ruptured rete MCA aneurysms in the literature is usually good. The majority of patients (15/17, 88.2%) presented a good recovery (GOS score 4 or 5), while 11.7% of patients (2/17) presented a bad recovery (GOS score 1-3).²

Discussion

Aneurysms associated with arterial twigs in the MCA (rete MCA) are a very rare finding, although a possible combination of fragile twigs, high hemodynamic stress, and congenital proneness certainly results in a higher risk of aneurysm formation and rupture.

There are five main types of MCA anomalies: accessory, duplicated, duplicate origin, fenestrated, and rete MCA. They appear to occur in a lower incidence than anomalies of other major intracranial arteries.³ Coined by Cho et al,¹ the term rete MCA refers to a vessel with a twig-like arterial network in the M1 segment,^{1,16} previously also described as twig-like MCA, unfused MCA, aplastic MCA, moyamoya phenomenon, moyamoya-like vessel, or anomalous collateral artery. It may originate from the original MCA trunk (i.e., T-MCA) or from an anomalous collateral artery (i.e., T-NACA), with a common finding of M1 atresia.²

This cerebrovascular anomaly appears to be more prevalent in Asians, and its incidence ranges from 0.11 to 1.17%.^{1,8,12} It may be a congenital anomaly, resulting from a regression interference of the fetal arterial network, or an acquired anomaly, resulting from an MCA steno-occlusion with collaterals formation. The similarity of the rete MCA with the fetal arterial network favors the congenital theory. Specifically for rete MCA extending from an anomalous collateral artery, the finding of a M1 atresia associated with a more caliber anomalous collateral artery, such as our case, also favors the congenital theory. Hemodynamic and wall shear stress are considered the main pathophysiological mechanism of aneurysm formation in the twig-like vessel.^{13,15} The adult MCA can relieve the pressure from the internal carotid artery, but the arterial twigs are usually thinner and the muscular layer is less developed, leading to structural fragility and flow-related aneurysms.² The main differential diagnoses of rete MCA are moyamoya disease and moyamoya syndrome.³ Differently from them, the rete MCA is unilateral, occurs exclusively in the M1 segment with a weblike appearance, and maintains the caliber/flow of the distal MCA segment.¹

The dichotomy of “clip or coil” is also an important matter for aneurysms associated with MCA anomalies, following the current tendency of endovascular embolization for intracranial aneurysms. To date, coiling has not been used for the treatment of rete MCA aneurysms, differently from aneurysms of other MCA anomalies, for which it presented results similar to open surgery.¹⁶ Approaches of endovascular

embolization have been attempted for rete MCA aneurysms, but failures occurred concerning the impeded advancement of the microcatheter (for gluing), with further conservative approach, and puncture of the lenticulostriate artery (for coiling), with further parent vessel sacrifice.^{2,6} The preference of open surgery for rete MCA aneurysms in the literature is justified by the prior technical constraint and aneurysm characteristics, such as unconventional location. Besides, the higher rate of ICH/IVH and tortuous twig angioarchitecture favors the use of open surgery. Nevertheless, the use of coiling may be a good alternative for feasible cases (e.g., no significant ICH/IVH, operator experience).

Conclusion

Although rare, it is important to be aware of and understand MCA anomalies and their concurrent vascular changes, such as rete MCA aneurysms. Aneurysms of rete MCA from anomalous collateral artery are a singular condition, with few cases in the literature. Microsurgical clipping is the main treatment procedure.

Authors' Contributions

Joaquim Francisco Cavalcante-Neto was involved in literature search, manuscript preparation, and manuscript editing and review. Gabriel de Almeida Monteiro contributed to literature search, manuscript preparation, and data analysis. Ariane Butke Brandt and Giovanna Esmeraldo Paz Soares helped in manuscript preparation and data analysis. Davi Jorge Fontoura Solla, Gerardo Cristino-Filho, Paulo Roberto Lacerda Leal, and Keven Ferreira da Ponte edited and reviewed the manuscript.

Conflict of Interest

None declared.

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