Clinical Case Report

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Duplicated middle cerebral artery origin with an aneurysm

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Abstract

Rationale: Middle cerebral artery (MCA) anomalies are relatively rare and often related to aneurysms. Familiarity with these anomalies is important in resolving problems that arise in the complex angioarchitecture. Reports often describe that aneurysms that are related to accessory or duplicated MCA are often located at its origin.

Patient concerns: A 59-year-old man presented with a headache for 10 days, without nausea and vomiting. The physical examination was negative.

Diagnosis: A computed tomography (CT) scan revealed an intracerebral hematoma in the deep right frontal lobe, near the caudate nucleus. Digital subtraction angiography (DSA) revealed an anomalous duplicated origin of the right MCA, with occlusion of the main MCA trunk as well as twisting and dilation of the accessory MCA trunk. A wide-necked aneurysm was located at a sharp curve of the tortuous accessory MCA trunk. A ruptured aneurysm related to a duplicated MCA origin was diagnosed.

Interventions: Open surgery was rejected by the patient; hence, palliative endovascular coil embolization of the larger daughter sac was performed.

Outcomes: The postoperative course was uneventful. There was no rebleeding at 8-months follow-up.

Lessons: MCA anomalies are relatively rare and often related to aneurysms. It is important to be familiar with these anomalies as related lesions often manifest within a complex angioarchitecture. Aneurysms at the trunk of an anomalous MCA are a rare entity and open surgery may be recommended.

Abbreviations: ACA = anterior cerebral artery, CT = computed tomography, DSA = digital subtraction angiography, ICA = internal carotid artery, MCA = middle cerebral artery.

Keywords: accessory middle cerebral artery, anatomic anomaly, cerebral aneurysm, duplicated origin, middle cerebral artery

1. Introduction

Intracranial arterial variations are routinely observed by radiologists, meanwhile, middle cerebral artery (MCA) anomalies are relatively rare. MCA anomalies are widely classified into varying types: accessory, duplicated, duplicated origin, fenestrated, or twig-like.[1] Among these, a duplicated MCA origin refers to a distal fusion of the duplicated MCA or accessory MCA, which occurs in 0.11% of cases.[2] Though rare, aneurysms may be associated with these MCA anomalies.[3–9] Aneurysms related to accessory or duplicated MCA are mostly located at the origin of the MCA, as reported. Herein, we report a case of a ruptured aneurysm related to a duplicated MCA origin, which arose from the trunk of the accessory MCA, which was accompanied by occlusion of the main MCA trunk.

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2. Case report

Ethical approval was obtained from the Human Research Ethics Committee of Tianjin Huanhu Hospital and the patient consent was obtained. A 59-year-old man presented with a headache for 10 days, without nausea and vomiting. The physical examination was negative. He had a history of hypertension for 6 years and tobacco use for 30 years. A computed tomography (CT) scan revealed an intracerebral hematoma in the deep right frontal lobe, near the caudate nucleus (Fig. 1A). Traditional digital subtraction angiography (DSA) revealed an aneurysm-like lesion at the right M1area with a lump of plexiform small vessels nearby along with a convoluted angioarchitecture (Fig. 1B). After carefully scrutiny of the three-dimensional rotational angiograph, the angioarchitecture was determined. In doing so, we found an anomalous duplicated origin of the right MCA, with occlusion of the main MCA trunk as well as twisting and dilation of the accessory MCA trunk. The accessory MCA trunk originated from the A1 to A2 conjunction of the anterior cerebral artery (ACA). A wide-necked aneurysm was located at a sharp curve of the tortuous accessory MCA trunk, with 2 daughter sacs (bleb) (Fig. 1D–G). A contralateral injection confirmed this result (Fig. 1C). Open surgery was rejected by the patient; hence, palliative endovascular coil embolization of the larger daughter sac was performed (Fig. 1H and I). The postoperative course was uneventful. There was no rebleeding at 8-months follow-up.

3. Discussion

Though intracranial arterial variations are a frequent finding, MCA anomalies are relatively rare. During embryonic development, the

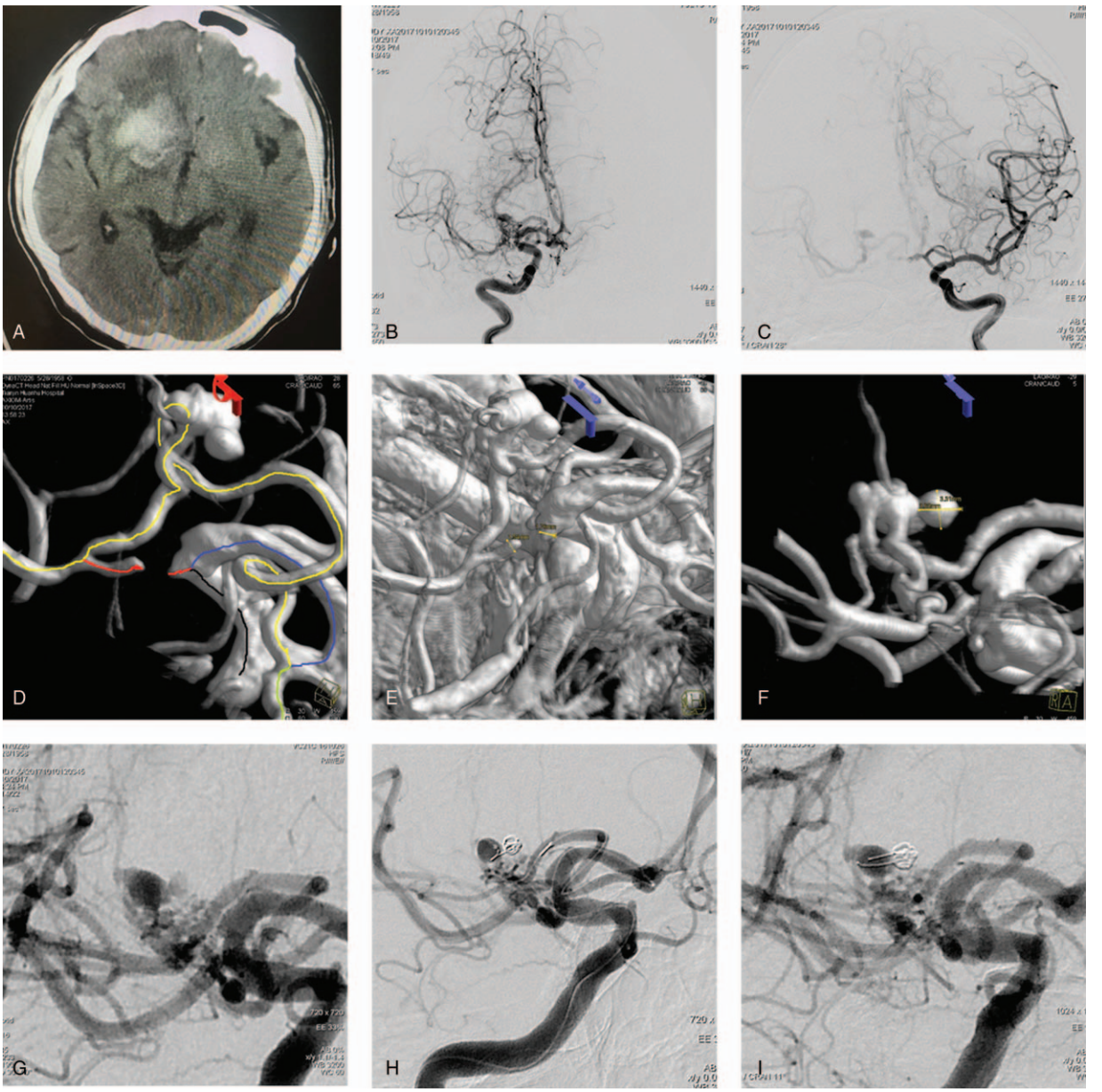

Figure 1. CT scan and emergency cerebral angiography of a 59-year-old man with a headache that persisted for 10 days. (A) CT scan showing an intracerebral hematoma in the deep right frontal lobe near the caudate. (B) Traditional DSA revealed an aneurysm-like lesion at the right M1 area, with plexiform small vessels nearby. (C) A contralateral injection showing the tortuous accessory MCA and the aneurysm. (D) Three-dimensional angioarchitecture showing the ICA (black line), A1 (blue line), and A2 (green line) segments of the ACA, the tortuous accessory MCA (yellow line), the occluded MCA (red line), and the aneurysm with 2 blebs. (E) Three-dimensional reconstruction of Dyna CT showing the occluded main truck of the MCA, the tortuous accessory MCA, and the aneurysm. (F) Three-dimensional reconstruction of rotational angiography showing daughter sacs of the aneurysm. (G) Injection from the operating angle showing the aneurysm and targeted sac. (H) Injection during the embolization showed twisted access (reflected by the micro catheter). (I) Final outcome of embolization. ACA = anterior cerebral artery, CT = computed tomography, DSA = digital subtraction angiography, ICA = internal carotid artery, MCA = middle cerebral artery.

distal portion of the internal carotid artery (ICA) bifurcates into cranial and caudal branches. The anterior cerebral artery (ACA) develops from the cranial branch. Moreover, the MCA develops after the ACA, which can be considered its collateral branch. Initially, multiple plexiform arterial twigs originate from the cranial branch. With development, these plexiform arterial twigs fuse and regress, forming lateral striate arteries and the normal trunk of the MCA (Fig. 2A). Interference in the process of fusion and regression results in anomalies of the MCA. Though controversial, MCA anomalies are commonly divided into five types: accessory (Fig. 2C), duplicated (Fig. 2F), duplicated origin (Fig. 2D and G), fenestrated (Fig. 2E), and twig-like (Fig. 2H). An accessory MCA, duplicated MCA, and duplicated MCA origin all exhibit double trunk originations. Therefore, differentiation between an accessory and duplicated MCA involves identification of the origin of the anomalous trunk, with the accessory MCA originating from the ACA and the duplicated MCA originating from the ICA. A duplicated MCA origin uniquely exhibits an angioarchitecture containing fusion of the distal portions of the duplicated or accessory MCA.[1] Rarely does the MCA present with 3 trunks: an accessory, duplicated, and normal trunk.[5] A duplicated MCA origin and fenestrated MCA both have a fenestrated structure. Double or single

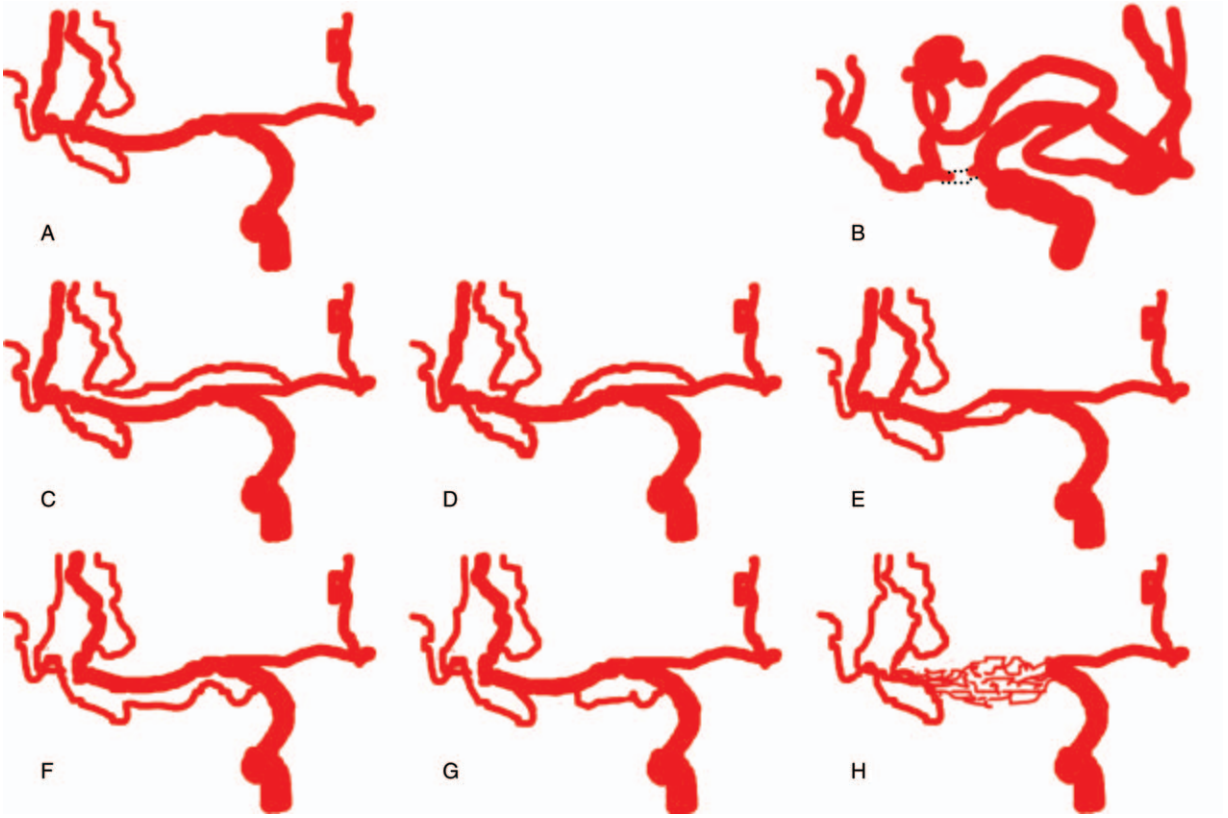

Figure 2. A diagram of MCA anomalies, normal MCA, and our case. (A) A normal MCA. (B) Our case showed a distal fusion of the accessory and main MCA (occluded, dotted line), according with the term duplicated origin of MCA. (C) The accessory MCA is restricted to the anomalous artery that arises from the ACA. (D) The duplicated origin of MCA with a distal fusion of the accessory and main MCA was showed. (E) MCA fenestration refers to a single truck from the ICA and a fenestrated structure. (F) The duplicated MCA refers to the anomalous artery arising from ICA. (G) The duplicated origin of MCA with a distal fusion of the duplicated and main MCA was showed. (H) The twig-like MCA refers to a plexiform arterial network exists instead of the normal MCA trunk, with normal cortical branches. ACA = anterior cerebral artery, ICA = internal carotid artery, MCA = middle cerebral artery.

origin MCA trunks facilitate the differentiation of the 2 aforementioned anomalies. A twig-like MCA exhibits an angioarchitecture in which a plexiform arterial network exists in place of the normal MCA trunk, but has normal cortical branches. Notably, atwig-like MCA is difficult to distinguish from unilateral moyamoya disease.[1,2]The observed vasculature structure in our case (Fig. 2B)is described as a duplicated origin of MCA with fusion of the distal portions of the accessory and main MCA trunks. Moreover, the main MCA trunk was occluded. Teramoto et al[7] reported a similar case with characteristics of a hematoma in the same region, an anomalous accessory MCA trunk from A1, a distal fusion of the double MCA trunks, an aneurysm at the trunk of the accessory MCA, and stenosis of the main trunk.

Although these collateral anomalous vessels may play an important role in supplying blood flow to the territory of the MCA, particularly in acute occlusion of the MCA, aneurysms have been reported relating to these anomalies.[3–10] Additional aneurysms located at other arteries are often found in these patients [11]; however, patients with aneurysms in anomalous MCA structures are at elevated risk of rupture.[11] Intracranial aneurysms in patients with a normal angioarchitecture often arise from bifurcation of vessels. This seems applicable to MCA anomalies. Most aneurysms associated with an accessory or duplicated MCA were reported at the origins of the anomalous vessel. Very few aneurysms in the trunk of an anomalous MCA have been reported.[3–5,7] Notably, abnormal vasculature morphologies and hemodynamic factors of the anomalous MCA may contribute to these aneurysms. As in our case, occlusion of the main MCA resulted in a compensatory dilatation of the accessory MCA. The degree of tortuosity and elasticity of the accessory MCA may result in a heavy flow burden, with a sharply curved course producing a high shearing force to the vessel wall at the dome of the curve. Both of these features result in a high risk of aneurismal formation and rupture.

Both endovascular embolization and open surgery have been reported for the treatment of aneurysms at the origins of the accessory or duplicated MCA, with open surgery being reported more frequently than endovascular embolization. Both treatment options achieve a good result. [8,9,12,13] Notably, aneurysms at the trunk of an anomalous MCA may be more appropriately treated by an open surgery than by endovascular treatment. Based upon the limited number of reports and our case, these aneurysms often have a wide neck, a sharply curved parent artery, and a tortuous access for catheterization (Fig. 1B–I), which might make endovascular treatment difficult and highly risky. In the reported cases of aneurysms at the trunk, all were treated by open surgery, except a dissecting aneurysm that was treated by endovascular glue embolization. All cases exhibited a good outcome. [3–5,7] As for our case, endovascular embolization of the larger daughter sac of the aneurysm was performed as a type of palliative treatment (Fig. 1I) due to the rejection of open surgery by the patient. There may be a high risk of aneurysm rupture in the

subsequent years, with open surgery being more appropriate for this patient.

4. Conclusion

MCA anomalies are relatively rare and often related to aneurysms. It is important to be familiar with these anomalies as related lesions often manifest within a complex angioarchitecture. Aneurysms at the trunk of an anomalous MCA are a rare entity and open surgery may be recommended.

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