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A Ruptured Aneurysm Located at a Collateral Artery That Extended from the Proximal A2 Segment to the M1 Segment, Associated with an Anomalous Branch of the Anterior Choroidal Artery and Middle Cerebral Artery Hypoplasia: Case Report

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Abstract

We here describe the first case of a ruptured aneurysm located at a collateral artery that extended from the proximal A2 segment to the M1 segment, which was associated with an anomalous branch of the anterior choroidal artery and middle cerebral artery (MCA) hypoplasia. The aneurysm was revealed by angiograms and intraoperative findings. No previous accounts have been published of such an extremely rare vessel anomaly. In practice, this case highlights the urgent need to preoperatively recognize such vascular anomalies, as well as to better understand the collateral blood supply in cerebral ischemia associated with these MCA anomalies. Such knowledge will be helpful for planning optimal surgical procedures.

Key words: anomaly, aneurysm, subarachnoid hemorrhage

Introduction

Three types of congenital vascular anomalies involving the middle cerebral artery (MCA) have been described, including accessory MCA (acc-MCA), duplicated MCA (dup-MCA), and fenestrated MCA anomalies.[1–3] In addition, Liu et al.[4] reported a rare abnormality of the MCA, which they named as twig-like MCA, which appeared to be a network between the bifurcation of the internal cerebral artery (ICA) and the insular segment of the MCA, and it was different from the collateral circulation due to acquired occlusion or moyamoya disease. Aside from these malformations, an anomalous collateral artery may develop in association with hypoplasia of the MCA to maintain the cerebral blood flow to the MCA area. In this report, we describe an extremely rare case of a ruptured aneurysm located at a collateral artery that extended from the proximal A2 segment to the M1 segment, which was associated with an anomalous branch of the anterior choroidal artery and MCA hypoplasia.

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Case Report

A 76-year-old male, who was previously treated for hypertension and myocardial infarction and had been taking antiplatelet drug for medication, suffered a sudden onset of headache and a loss of consciousness. He was transferred to our hospital. On admission, he was comatose, and had a Glasgow Coma Scale of E1V2M5 with right hemiparesis. A three-dimensional computed tomography angiograph (3D-CTA) showed left MCA hypoplasia and an anomalous branch of the anterior choroidal artery (telencephalic branch), which shared in supplying the left MCA territory, and no obvious aneurysmal shadow (Fig. 1B, E). Serial digital subcutaneous angiography (DSA) revealed an aneurysmal shadow located at a collateral artery that extended from the proximal A2 segment to the M1 segment (Fig. 1C, E; *arrowhead* and *arrows*). A follow-up 3D-CTA showed the growth of the aneurysm (Fig. 1D). A schematic illustration showed the angiographical findings in detail (Fig. 1E). The interval change of the aneurysmal shadow raised concern for a ruptured aneurysm. We planned direct neck clipping or trapping from a pterional approach. A left frontotemporal

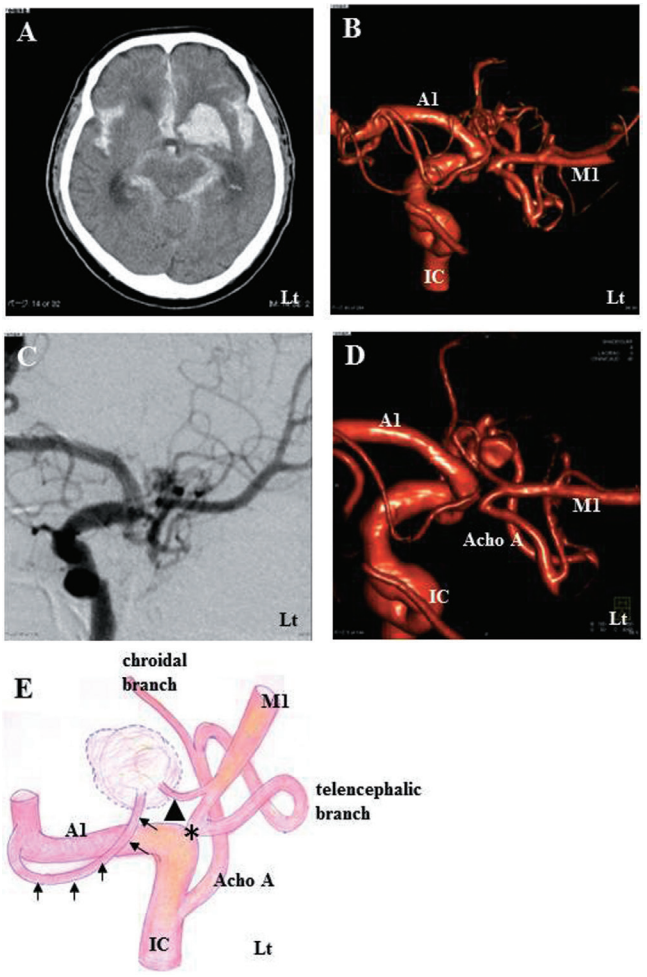

Fig. 1 A: A computed tomography (CT) scan on admission revealed a diffuse subarachnoid hemorrhage with an intracerebral hematoma in the left putaminal area. B: A three-dimensional CT angiogram (3D-CTA) showed left middle cerebral artery (MCA) hypoplasia and an anomalous branch of the anterior choroidal artery (telencephalic branch), which shared in supplying the left MCA territory, and no obvious aneurysmal shadow. C: Serial digital subcutaneous angiography showed an aneurismal shadow located at the collateral artery that extended from the proximal A2 segment to the M1 segment. D: Follow-up 3D-CTA showed the growth of the aneurysm. E: A schematic illustration showed the angiographical findings in detail (*arrows*: a collateral artery that extended from the proximal A2 segment to the aneurysm, *arrowhead*: a collateral artery that extended from the proximal A1 segment to the aneurysm, *asterisk*: the left MCA hypoplasia). Acho: anterior choroidal artery, Lt: left.

craniotomy was performed about one month after ictus. After the dissection of the carotid cistern, we traced the left ICA (Fig. 2A), and observed that the left MCA hypoplasia (Fig. 2A, C, D, F, J; *asterisk*) with an anomalous branch of the anterior choroidal artery (telencephalic branch) shared the left MCA territory. In addition, the collateral artery that extended from the proximal A2 segment to the M1 segment could be viewed (Fig. 2E, F; *arrowhead*), and was traced toward the aneurysm (Fig. 2C, G, J; *arrows*). With further retraction of the frontal lobe, the encapsulated old clot hematoma, including an aneurysm, was confirmed (Fig. 2H, J; *dotted line*). The area around the encapsulated hematoma was completely dissected and trapped successfully (Fig. 2J; *black line*). Histological examination showed that the red sac was quite different from the true aneurysm. The aneurysmal wall consisted of a fibrin layer, which was laminated and intermingled with blood clot elements (Fig. 3) without inner reticular layer. These histological features indicated that the portion of the excised lesion was a pseudoaneurysm. The patient's postoperative course was uneventful with no vasospasms or cerebral infarction.

Discussion

Intracranial vascular anomalies involving the MCA are relatively rare. Teal et al.[1] established a distinction between two types of accessory MCA. The presence of vessels arising from the distal ICA between the anterior choroidal artery and the terminal bifurcation of the ICA and feeding the vascular territory of the normal MCA is referred to as a dup-MCA. On the other hand, a vessel that originates between the A1 and proximal A2 segment of the anterior cerebral artery (ACA), reaches the sylvian fissure and feeds the territory of the MCA, is defined as an acc-MCA.[1,3] Liu et al.[4] reported a rare abnormality of the MCA, which they named as twig-like MCA, which appeared to be a network between the bifurcation of the ICA and the insular segment of the MCA, and this was different from the collateral circulation arising due to acquired occlusion or moyamoya disease. Yamamoto et al.[5] suggested that while an acc-MCA is a true anomalous artery, a dup-MCA is instead a variation in the branching of the ICA. In our case, the anomalous collateral artery at the branching of the anterior choridal artery may have developed in association with hypoplasia of the MCA to maintain the cerebral blood flow to the MCA area. The embryological explanation for anomalies and variations of the MCA remains unclear. The MCA develops after the ACA, and the ACA is considered a continuation of the primitive ICA. Thus, the MCA can be regarded as a branch of the ACA.[6] Embryologically, the MCA can be recognized in a 7 to 12 mm embryo as twigs sprouting from the ICA proximal to the ACA. By the 16 to 18 mm stage, the MCA has become more prominent and supplies branches that spread over the cerebral hemisphere.

The associations between a dup-MCA, acc-MCA, twig-like MCA, and cerebral aneurysms have been well-documented.[4,7,8] However, it is not clear whether these associations are a chance occurrence or whether they are related by an unknown mechanism. In practice, it is important to preoperatively recognize such vascular

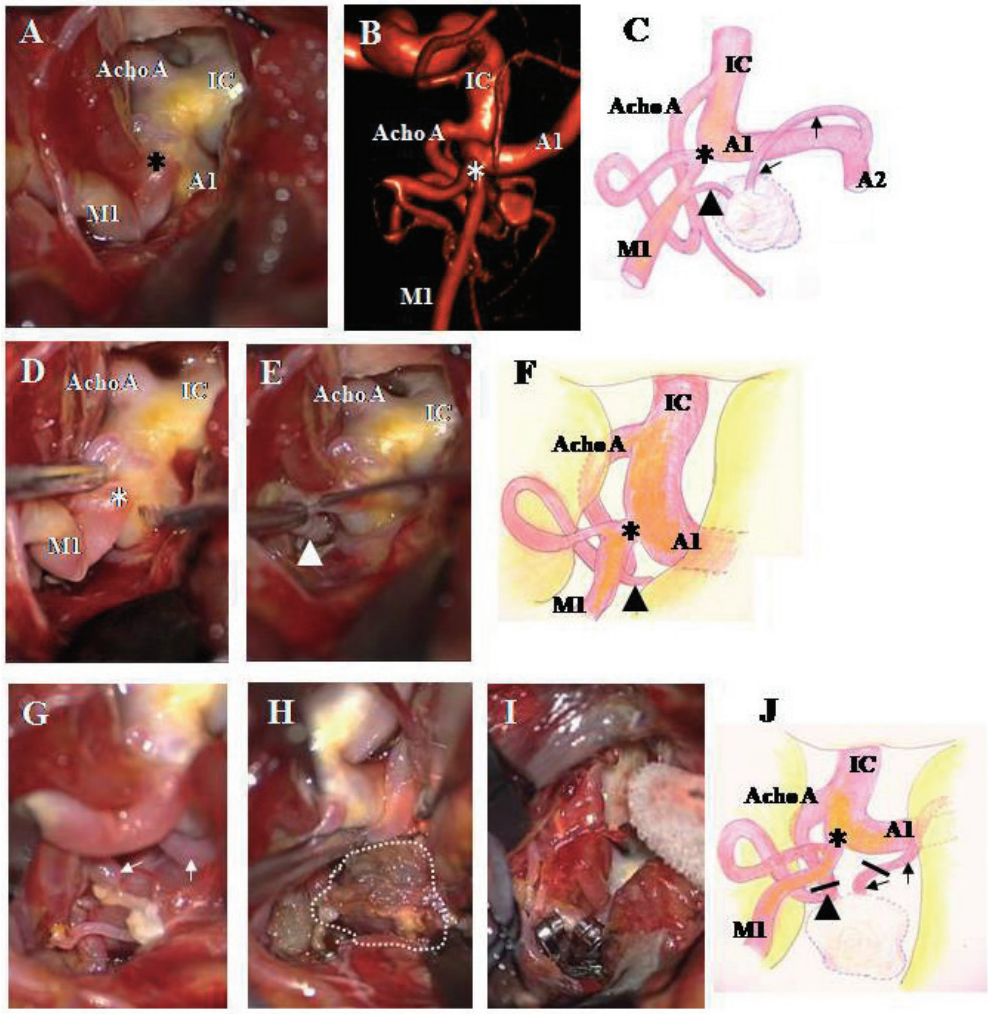

Fig. 2 A left frontotemporal craniotomy was performed. An intra-operative photograph and three dimensional computed tomography angiogram showed the left ICA and the MCA hypoplasia with an anomalous branch of the anterior choroidal artery (Acho), which shared the left MCA territory (A, B, D; *asterisk*). After the dissection of the chiasmatic cistern, we confirmed the abnormal vessels from the M1 to the aneurysmal shadow (E; *arrowhead*), traced the ACA, and observed the collateral artery that extended from the proximal A2 segment to the M1 segment (G; *arrows*). With the further retraction of the frontal lobe, we were able to trace the abnormal artery to reach the encapsulated old clot hematoma, including the aneurysm (H; *dotted line*). The area around the encapsulated hematoma was completely dissected and thereafter trapped successfully (I). A schematic illustration showed the intraoperative findings in detail (C, F, J; *arrow*: a collateral artery that extended from the proximal A2 segment to the aneurysm, *arrowhead*: a collateral artery that extended from the proximal A1 segment to the aneurysm, *asterisk*: the left MCA hypoplasia, *dotted line*: the encapsulated old clot hematoma, including the aneurysm). ACA: anterior cerebral artery, ICA: internal cerebral artery, MCA: middle cerebral artery.

anomalies so that the best surgical procedures for treating a cerebral aneurysm can be selected, since the collateral blood supply in patients with cerebral ischemia is associated with the presence of these MCA anomalies. With respect to their clinical importance, the MCA anomalies have the potential to serve as a collateral blood supply to the MCA territory in cases of MCA occlusion. An MCA anomaly may also play an important role in supplying collateral blood flow to the frontal lobe and basal ganglia through the perforating arteries. The MCA anomaly can be a form of collateral circulation to the anterior frontal lobe, but it cannot supply flow with sufficient power to the main MCA territory.[9] Similarly, an MCA anomaly can be collateral to the anterior temporal lobe, but it does not seem to supply enough blood to the main MCA territory. Komiyama et al.[10] reported that the dup-MCAs have perforating arteries that attach them to the anterior perforated substance. Therefore, it is generally accepted that special attention should be paid to avoid ischemic complications in patients with these anomalies, and to

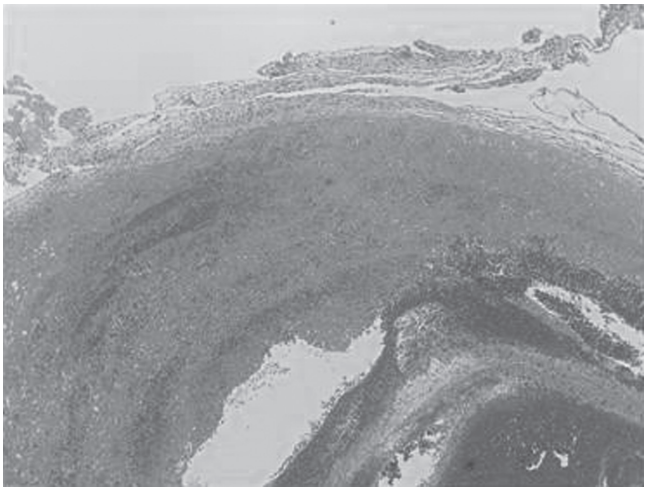

Fig. 3 Photomicrograph of the excised pseudoaneurysm showed the aneurysmal wall consisted of a fibrin layer, which was laminated and intermingled with blood clot elements. Fresh and old thrombus were adherent to the inner surface of the pseudoaneurysmal wall.

find out whether a temporary clip should be used to disrupt the blood flow and thus control the potential of a premature rupture.

In this case, the ruptured aneurysm was located at a collateral artery that extended from the proximal A2 segment to the M1 segment with MCA hypoplasia. Han et al.[7] reported a case in which an acc-MCA was associated with MCA aplasia. Their surgical exploration revealed a cordlike rudimentary structure, which arose at the ICA bifurcation and an artery that originated at the proximal A1 segment and coursed into the sylvian fissure. Such collateral circulation can be developed after MCA occlusion, which usually results from leptomeningeal connections from the ACA to the branch of the MCA. In our case, trapping with complete sacrifice of the encapsulated hematoma, including the aneurysm, was performed without any neurological deficit or infarction on the follow-up CT after surgery. Intraoperative findings showed no perforators observed surrounding the encapsulated hematoma, which suggested that the perforators may not be connected with these abnormal lesions. Therefore, the perforators located in each side of the trapped vessel will be fully perfused from the proximal A2 segment or the M1 segment. In addition, the sacrificed collateral artery that extended from the A2 segment to the M1 segment was similar size with an anomalous branch of the anterior choroidal artery (telencephalic branch), which these both vessels shared the left MCA territory. Accordingly, the trapping of the aneurysm located on the collateral artery that extended from the A2 segment to the M1 segment might induce cerebral blood flow reduction. Therefore, bypass procedure and intraoperative monitoring should be prepared to perform the trapping of the vessels safely.

In terms of the surgical procedure, proximally securing the aneurysm is extremely important to prevent intraoperative rupture. In this case, we had to dissect and confirm the ICA and trace the abnormal branch of the anterior choroidal artery to control the intraoperative bleeding. Therefore, it is generally accepted that special attention should be paid to avoid ischemic complications, and to find out whether a temporary clip should be used to disrupt the blood flow, and thus control the possibility of premature rupture.

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