Online October 9, 2021
Journal of Neuroendovascular Therapy 2022; 16: 376–380
DOI: 10.5797/jnet.cr.2021-0079

Case Report

A Case of Aneurysmal Subarachnoid Hemorrhage with Middle Cerebral Artery Aplasia at 30 Weeks of Pregnancy

Yuta Oi, Yasuo Inoue, Ichita Taniyama, Mitsuru Shirato, and Takashi Houri

Objective: We report a case of subarachnoid hemorrhage (SAH) that occurred at 30 weeks of pregnancy and was treated by coil embolization in a woman with middle cerebral artery (MCA) aplasia.

Case Presentation: A 40-year-old woman who was 30 weeks pregnant presented to the emergency department with a half-day history of headache and nausea. She had sudden onset headache and her symptom did not improve. There was no neurological deficit. Head CT at the referring hospital revealed SAH. The fetal state was stable. There was no sign of threatened premature delivery. Head MRA revealed aplasia of the left MCA and aneurysm with a daughter sac at the A1 segment of the anterior cerebral artery (ACA). Head DSA revealed that the A1 aneurysm with a daughter sac arose from the anomalous collateral artery leading to a plexiform network. The diagnosis was SAH due to rupture of an A1 aneurysm. Performance of less invasive coil embolization seemed to be possible and was carried out under general anesthesia. The operation was completed after placing one coil and confirming that most of the aneurysmal dome was embolized, including the daughter sac. There was no cerebral vasospasm and no obvious neurological deficit. Antiplatelet drugs were only required for 10 days after the operation. Pregnancy was stable and the patient delivered a baby by cesarean section at 38 weeks of pregnancy.

Conclusion: A rare case of aneurysmal SAH in a pregnant woman with MCA aplasia was successfully treated by endovascular surgery.

Keywords subarachnoid hemorrhage, pregnancy, middle cerebral artery aplasia, rete middle cerebral artery, endovascular treatment

Introduction

Non-traumatic subarachnoid hemorrhage (SAH) is known as the main cause of stroke. SAH accounts for about 10% of all strokes.[1] The most common cause of SAH is rupture of intracranial aneurysm. The overall incidence of SAH was reported to be approximately 9 per 100000 person-years.[2] The incidence of SAH in Japanese populations is estimated to be approximately 22.7 per 100000 persons.[2] Recently, the incidence of SAH occurring during pregnancy is on the upward trend.[3] The reported frequency of SAH in pregnant woman ranges from 8 to 31 per 100000 deliveries.[3] We report a case of a pregnant woman who presented with SAH. This case was rare from the aspect of aneurysmal SAH in pregnant woman with aplasia of left middle cerebral artery (MCA). We report it along with the relevant literature review.

Department of Neurosurgery, Maizuru Medical Center, Maizuru, Kyoto, Japan

Received: June 11, 2021; Accepted: September 22, 2021
Corresponding author: Yuta Oi. Department of Neurosurgery, Maizuru Medical Center, 2410, Yukinaga, Maizuru, Kyoto 625-8502, Japan
Email: m10015yo@gmail.com

Case Presentation

A 40-year-old woman in her 30th week of pregnancy was admitted with a half-day history of severe headache and nausea. She had sudden onset headache in bathroom and her symptom did not improve. She had pregnancy-induced hypertension and her grandfather died of SAH. There was no neurological deficit, World Federation of Neurosurgical Societies (WFNS) grade II. CT at the referring hospital revealed SAH in the left Sylvian fissure (**Fig. 1**). Fetal state was stable. There was no sign of threatened premature

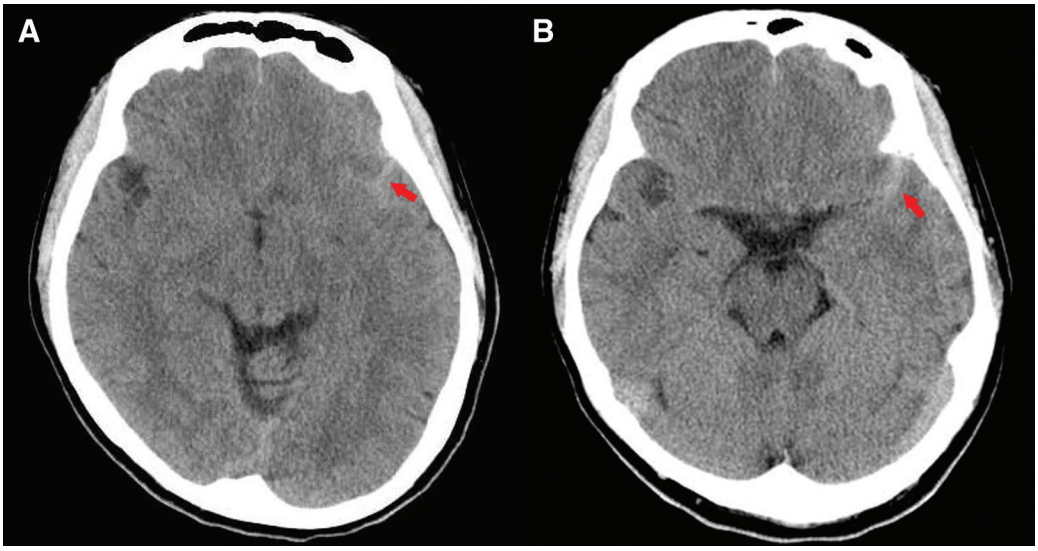

Fig. 1 CT at the referring hospital showing SAH (arrow) in the left Sylvian fissure. SAH: subarachnoid hemorrhage

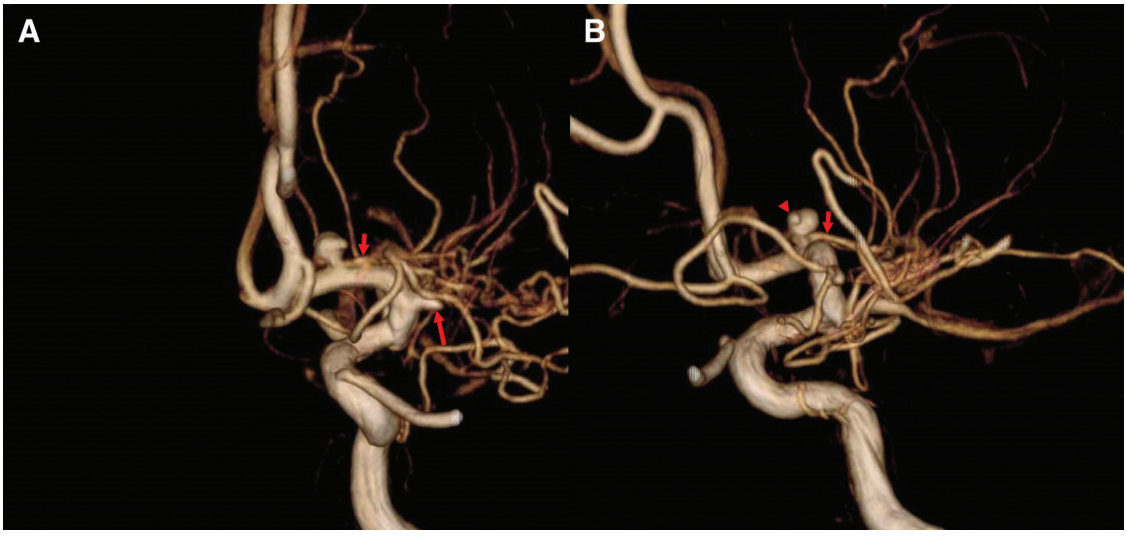

Fig. 2 Preprocedural angiogram showing aplasia of the left MCA and saccular left aneurysm at the bifurcation of the anomalous collateral artery and A1 segment of the ACA (**A**). Preprocedural angiogram showing a daughter sac at the dome of the aneurysm (**B**). (arrowheads: daughter sac; arrows: anomalous collateral artery from the A1 segment; long arrow: MCA aplasia) ACA: anterior cerebral artery; MCA: middle cerebral artery

delivery. She was transferred to our institution for further management.

MRA of head revealed aplasia of left MCA and aneurysm (neck 1.3 mm, dome 4.1 mm, and height 3.5 mm) with daughter sac at A1 segment of anterior cerebral artery (ACA). Diagnosis revealed SAH due to rupture of A1 aneurysm.

The initial left internal carotid artery angiogram revealed the A1 aneurysm with daughter sac arising from an anomalous collateral artery leading to a plexiform network (**Fig. 2**). The normal MCA was occluded in the M1 proximal portion. M1 perforating branches were present and a few lenticulostriate arteries were perfused from the anomalous collateral artery. The diameter of the MCA cortical branch at the neck of the A1 aneurysm was 0.73 mm, and this branch was the same as the anomalous collateral artery leading to MCA territory. The timing of imaging was the early stage of the arterial phase. The recurrent artery of Heubner arose from A2 and formed a perforating branch to MCA territory. It seemed to be possible to perform less invasive coil embolization. The endovascular procedure was performed under general anesthesia on the

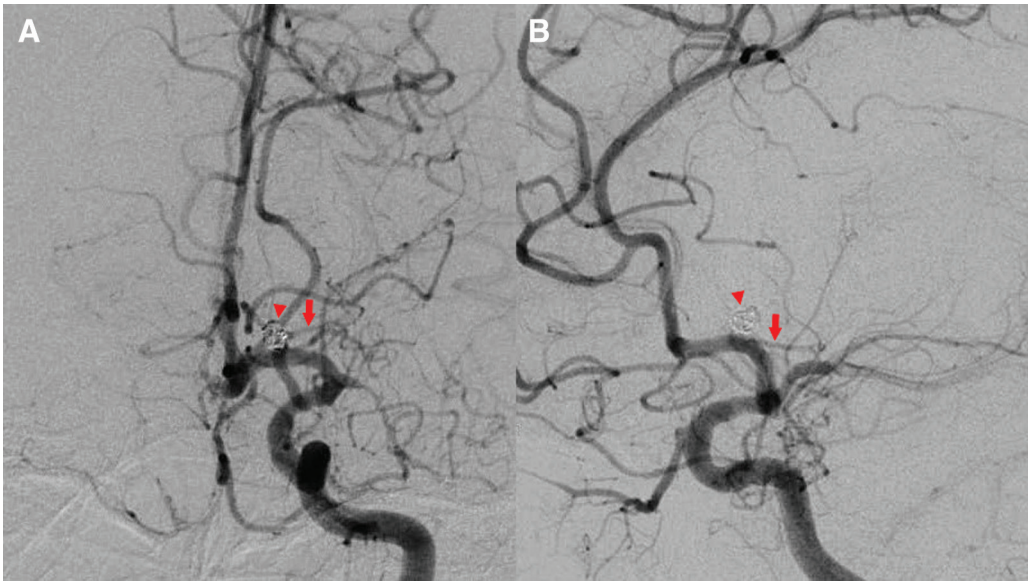

Fig. 3 Postprocedural angiogram showing that most of the aneurysmal dome including the daughter sac was embolized. (**A**) AP. (**B**) LAT. (arrowheads: aneurysm after coiling; arrows: anomalous collateral artery from the A1 segment)
AP: anteroposterior; LAT: lateral

same day. We used 6Fr Roadmaster (Goodman, Aichi, Japan) as guiding catheter. We performed 3D rotational angiography to determine the working angle, and this angle was used to separate the aneurysm and the branching arteries sufficiently (**Figs. 2**–**4**). We easily introduced an Excelsior SL-10 preshaped 45 microcatheter (Stryker, Kalamazoo, MI, USA) with a CHIKAI 14 microguidewire (Asahi Intecc, Aichi, Japan) to the neck of aneurysm and deployed a Target 360 Soft 3.5 × 10 coil (Stryker). A further attempt to place another coil was not successful. We considered that most of the aneurysmal dome including the daughter sac was embolized and then the operation was completed (**Fig. 3**). A subsequent CT showed no intracranial bleeding during the procedure and no hydrocephalus. Also, there was no old ischemic or hemorrhagic lesion in the left hemisphere. We continued heparinization during endovascular procedure and naturally reversed it. The patient was recovered from the anesthesia with no neurological deficit and transferred to the stroke care unit (SCU) for monitoring.

Immediate postprocedural physical and ultrasound examination of fetus did not show any abnormalities. The patient had no complications after the procedure and discharged on the 17th day with 10-day course of an antiplatelet therapy. The patient delivered a healthy baby with an emergent cesarean section on 38th week of gestation. Follow-up MRA at 32 months showed no coil compaction or regrowth of the aneurysm. Blood flow to the aneurysmal neck was scarcely noticeable on the MRA image. The patient is being regularly reviewed in our outpatient clinic.

Discussion

Differential diagnosis is required in a case with acquired MCA occlusion. In our case, arteriosclerotic occlusion was unlikely because the patient was a young woman. Arterial dissection was also unlikely because there was no mural hematoma or imaging finding of past stroke, and vasculitis was not suspected because there was no elevation of inflammatory markers (C-reactive protein and neutrophils). There were also no steno-occlusive changes of the internal carotid artery terminus and posterior cerebral artery, or transdural anastomosis. In addition, the lesion was unilateral. Given these facts, suspected spontaneous MCA aplasia was more likely than moyamoya disease.

An unusual type of vascular lesion of the MCA appears to be associated with occurrence of an aneurysm.[4] In the case reported here, DSA showed a plexiform network of an anomalous collateral artery from the A1 segment (**Fig. 4**). This is likely to reflect development of collateral blood flow associated with the occlusion. Hemodynamic stress is considered to be the main cause of aneurysmal formation.[5,6] According to Liu et al., a rete MCA is more fragile and less elastic than the normal MCA because it is usually thinner and the muscular layer is less developed.[6] Therefore, flow-related aneurysms are more easily formed.[4]

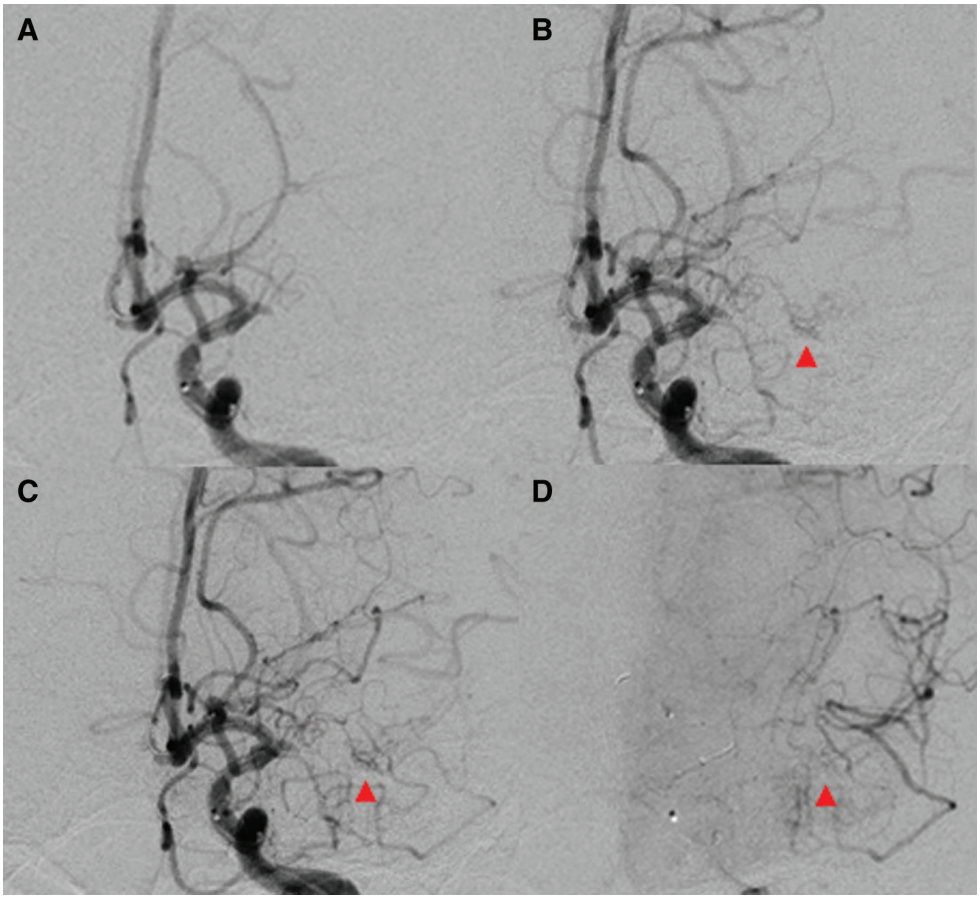

Fig. 4 Preprocedural angiogram showing perfusion to the left hemisphere in order from (**A**) to (**D**). (arrowheads: plexiform network of an anomalous collateral artery from the A1 segment)

MCA aplasia in our case did not strictly meet the definition of rete MCA or twig-like MCA,[4,7] but the anomalous collateral artery leading to MCA territory via the plexiform network was susceptible to hemodynamic stress. As a result, the aneurysm could have formed at the bifurcation of A1 and the anomalous collateral artery leading to MCA territory via the plexiform network.

It is unclear whether surgical clipping or endovascular coiling is the better procedure for therapeutic intervention for aneurysmal SAH in a pregnant woman. This case was rare from the perspective of aplasia of the left MCA that occurred during pregnancy. Considering this specific anatomy, vascular structure, we selected endovascular coiling. There is a similar case report of endovascular treatment of an A1 aneurysm with MCA anomaly.[8] The aneurysm had a wide neck, but it was successfully treated with a balloon remodeling technique. The choice of treatment for ruptured aneurysms should be comprehensively decided depending on the size, shape, and location of aneurysms. In this case, we judged this small aneurysm with narrow neck at bifurcation of A1 and the anomalous collateral artery leading to MCA territory via a plexiform network could be easily embolized without adjunctive technique in a relatively short time.

Successful endovascular treatment for aneurysmal SAH during pregnancy has been reported. However, there are several controversial issues: radiation exposure, contrast agent, anticoagulation, and antiplatelet drugs. First, a study reported that if irradiation to reproductive gland is below 50 mGy, the risk of fetal teratogenicity, growth retardation, and death does not increase.[9] Also, the irradiation risk to the fetus in coil embolization during pregnancy was assessed by a phantom study.[10] The absorbed fetal dose ranged from 0.17 to 2.8 mGy, during 15–45 min irradiation to the head of pregnant woman.[11] It is far below the risk of hereditary disease at birth or the natural cumulative risk of fatal childhood cancer by 15 years of age.[11] In this case, we used a 0.25 mmPb protector on the abdomen after guiding the catheter beyond the level of the renal artery. The maximum estimated absorbed fetal dose

was about 5.7 mGy (maximum radiation of 17.1 mGy/min and an abdomen exposure time of <20 s). Exposure to scattered radiation could not be avoided completely, but this was close to zero due to use of the protector and the long distance between the head and abdomen. It is low enough for fetus to avoid health hazard. Next, although it is well known that contrast agent reaches fetus through placenta, safe amount to use is still unclear. According to a case report, it was safe for maternal and fetal body to use contrast agent (200 ml).[12] In this case, we used 70 ml of contrast agent and did not see any problem. Finally, endovascular surgery required heparin-induced systemic anticoagulation to reduce the thromboembolic complications during the procedure. The usage of both heparin and antiplatelet agents increases the hemorrhagic risk in case of intraoperative precipitous labor or emergent cesarean section. In this case, we used aspirin (100 mg), cilostazol (200 mg), heparin (6000 units), and ozagrel (80 mg) on the operation day. After the surgery, we used only clopidogrel (75 mg) for 10 days. No hemorrhagic complication occurred. In Japanese guidelines, aspirin and cilostazol are contraindicated after 12 weeks, and clopidogrel and ozagrel can be used in pregnant women only if beneficial. Regarding the risk of ischemic complications, premedication with a single antiplatelet drug for coiling is widely accepted. Also, antiplatelet therapy might be a risk for a patient in mid-pregnancy because of the possible need for emergency caesarean section. In this case, the branching artery arose at the dome of the aneurysm and there was a strong risk of occlusion if the aneurysm was tightly packed. Thus, we used dual antiplatelet agents in this patient.

Cases of endovascular surgery are increasing recently and postoperative management is different according to the therapeutic technique. In this case, the aneurysm was small with narrow neck and so required no adjunctive technique as balloon neck plasty or stenting. In cases requiring no stent, we can finish the procedure more shortly without long-term postoperative antiplatelet drugs.

Conclusion

We experienced a rare case of aneurysmal SAH in a pregnant woman with MCA aplasia. This case was successfully treated by endovascular surgery. In some cases, endovascular treatment for aneurysmal SAH of pregnant woman is useful. We have to consider several factors in advance, such as pregnant state, estimated delivery date, difficulty of endovascular procedure, and medication after treatment.

Disclosure Statement

The authors declare that they have no conflicts of interest.

References

1) Toossi S, Moheet AM. Intracerebral hemorrhage in women: a review with special attention to pregnancy and the post-partum period. *Neurocrit Care* 2019; 31: 390–398.
2) de Rooij NK, Linn FH, van der Plas JA, et al. Incidence of subarachnoid haemorrhage: a systematic review with emphasis on region, age, gender and time trends. *J Neurol Neurosurg Psychiatry* 2007; 78: 1365–1372.
3) Limaye K, Patel A, Dave M, et al. Secular increases in spontaneous subarachnoid hemorrhage during pregnancy: a nationwide sample analysis. *J Stroke Cerebrovasc Dis* 2019; 28: 1141–1148.
4) Cho KC, Kim JJ, Jang CK, et al. Rete middle cerebral artery anomalies: a unifying name, case series, and literature review. *J Neurosurg* 2018; 131: 453–461.
5) Shin HS, Lee SH, Ryu CW, et al. Flow-related intracranial aneurysms associated with unfused arterial twigs relevant to different vascular anomalies: embryologic and hemodynamic considerations. *Acta Neurochir (Wien)* 2014; 156: 1637–1646.
6) Liu HM, Lai DM, Tu YK, et al. Aneurysms in twig-like middle cerebral artery. *Cerebrovasc Dis* 2005; 20: 1–5.
7) Ota T, Komiyama M. Twig-like middle cerebral artery: embryological persistence or secondary consequences? *Interv Neuroradiol* 2021; 27: 584–587.
8) Cekirge HS, Peynircioglu B, Saatci I. Endovascular treatment of an "anterior cerebral artery" aneurysm in a patient with "embryonic unfused middle cerebral artery" anomaly: a case report. *Neuroradiology* 2005; 47: 690–694.
9) Brent RL. The effect of embryonic and fetal exposure to X-ray, microwaves, and ultrasound: counseling the pregnant and nonpregnant patient about these risks. *Semin Oncol* 1989; 16: 347–368.
10) Marshman LA, Aspoas AR, Rai MS, et al. The implications of ISAT and ISUIA for the management of cerebral aneurysms during pregnancy. *Neurosurg Rev* 2007; 30: 177–180, discussion 180.
11) Marshman LA, Rai MS, Aspoas AR. Comment to "Endovascular treatment of ruptured intracranial aneurysms during pregnancy: report of three cases". *Arch Gynecol Obstet* 2005; 272: 93.
12) Takeuchi A, Sasaki U, Takeda T, et al. Ruptured intracranial aneurysm treated by embolization with GDC in pregnancy. *Jpn Cong Neurol Surg* 2004; 13: 533–538. (in Japanese)