

Case Report

Surgical removal of an arteriovenous malformation in the anterior perforated substance in a pregnant woman

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Abstract

Background: A tailor-made treatment is often required in arteriovenous malformations (AVMs) depending on the individual situation. In most cases, treatment strategy is usually determined according to the patient's Spetzler–Martin grade. However, in the present case, we were not able to treat the patient following the usual guidelines because of neurological symptoms and pregnancy.

Case Description: We describe a rare case of a 31-year-old woman in the 15th week of gestation who presented with an AVM in the anterior perforated substance (APS). She suffered a sudden coma and hemiplegia. A computed tomographic scan showed an enhanced mass and a huge hematoma in the basal ganglia and temporal lobe. The hematoma was successfully evacuated in an endoscopic procedure. Angiography showed that a 25-mm nidus in the APS was fed by the anterior choroidal arteries (AChAs) and the lenticulostriate arteries (LSAs). Therefore, we attempted to remove the nidus because the patient became alert with mild aphasia and hemiparesis 10 days after hemorrhage. The feeding arteries were cut under motor evoked potential (MEP) monitoring, and the nidus was totally resected leaving two of four AChAs and a single artery with several LSAs. The postoperative course was uneventful, and she gave birth to a healthy baby by caesarian delivery 122 days after the hemorrhage with only minor sequelae.

Conclusions: Surgical strategy with a device-administered anesthesia are suitable for removing large AVMs even in pregnant women and for the successful outcome of their pregnancies. Even after recovering from a coma and hemiplegia, MEP monitoring is effective for removing large AVMs even when located in the APS.

Key Words: Anterior perforated substance, arteriovenous malformation, motor evoked potential, pregnancy

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INTRODUCTION

An American Heart Association (AHA) Scientific Statement described the management of arteriovenous malformations (AVMs) in 2001 as follows:^[12] Surgical removal is strongly recommended for Spetzler–Martin (SM) grades 1 or 2 AVMs. For SM grade 3 AVMs, radiosurgery is recommended for those in

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deep locations or removal after endovascular embolization of large AVMs. In SM grades 4 or 5 AVMs, direct surgical treatment is not recommended.

Surgical extirpation of AVMs located in the anterior perforated substance (APS) is challenging because the surgical field is deep and vital arteries, such as the anterior choroidal arteries (AChAs) and the lenticulostriate arteries (LSAs), are in and around the nidus. The morbidity rate in the surgical treatment of basal ganglia AVMs is not low but 10–29%.^[14] Therefore, radiosurgery is advantageous in treating difficult AVMs such as deep-seated AVMs, as suggested in the AHA statement.^[10,12] However, surgeons usually have to wait for more than 1 year to obtain a cure. It is not reasonable to suggest radiosurgery for pregnant women because the risk of hemorrhage remains up to and through the time of the birth. Here, we describe a rare surgical case of an SM grade 3 AVM in the APS in a pregnant woman.

CASE REPORT

A 31-year-old woman in the 15th week of gestation suddenly lost consciousness and was transferred to our institution. She was in a deep coma with her left pupil dilated and right hemiplegia. Computed tomographic scans showed an intracerebral hemorrhage in the left basal ganglia extending to the temporal lobe and a small, enhanced mass above the APS [Figure 1a]. An endoscopic hematoma evacuation was promptly performed [Figure 1b]. She recovered consciousness and motor function soon after the operation. The fetal growth and development was also satisfactory. Angiograms showed a nidus, fed by the AChAs, the LSAs, and perforators from the A1 segment of the anterior cerebral artery, which drained into the basal vein of Rosenthal (BVR), the deep middle cerebral vein, and the anterior communicating vein. The maximum diameter of the nidus was 25 mm with an SM grade of 3 [Figure 1c and d]. Curative surgery was indicated because both she and her husband wanted her pregnancy to be continued to full term.

A bolus injection of rocuronium bromide (0.9 mg/kg) and propofol (2 mg/kg) was given before tracheal intubation. She received a continuous infusion of propofol (3–5 mg/kg/h) thereafter. An obstetrician monitored the fetal heartbeats. We performed an orbitozygomatic craniotomy and a partial temporal lobectomy to obtain a wide surgical field. A grid strip with 16 electrodes (Unique Medical, Tokyo, Japan) was inserted into the subdural space to facilitate electrical stimulation to the motor cortex (Neuropack X1, Nihon Kodens, Tokyo, Japan). The filters were at 20 Hz and 3 kHz (low- and high-band passes, respectively). We confirmed the muscle action potentials from the contralateral thenar muscles. Intraoperatively, the motor cortex was stimulated at 2 mA above the threshold level. A monopolar anodal electrical stimulus with five pulses was applied. The frequency of the train pulse was 500 Hz, and the duration of each single pulse was 200 ms.

The internal carotid artery, the A1 segment of the anterior cerebral artery, and the M1 segment of the middle cerebral artery were all well exposed after widely opening the Sylvian fissure. More than 10 perforating arteries were found to feed the nidus in the APS adjacent to the M1 segment. There were four AChAs feeding the nidus, and the distal two were cut because the test occlusion for 5 min did not induce a depression of the motor evoked potential (MEP) [Figure 2a]. The proximal two AChAs were preserved because the amplitude decreased 1 min after clamping [Figure 2b]. A few A1 perforators were also cut after the test occlusion. The LSAs were clamped, one at a time, proximally to distally, with Sugita microclips (Mizuho Ika, Tokyo, Japan) and the MEP was measured at 1 and 5 min after the occlusion. The LSAs were cut when the amplitude was not decreased [Figure 2c]. An LSA branching from the most distal part of the M1 segment was preserved because the amplitude decreased 5 min after clamping [Figure 2d], even though there was no MEP depression at 1 min. The preserved LSA was revealed passing through the nidus [Figure 3a]. The nidus was lifted from the APS after all the arteries supplying the nidus were cut [Figure 3b]. We confirmed that the nidus was not stained by indocyanine

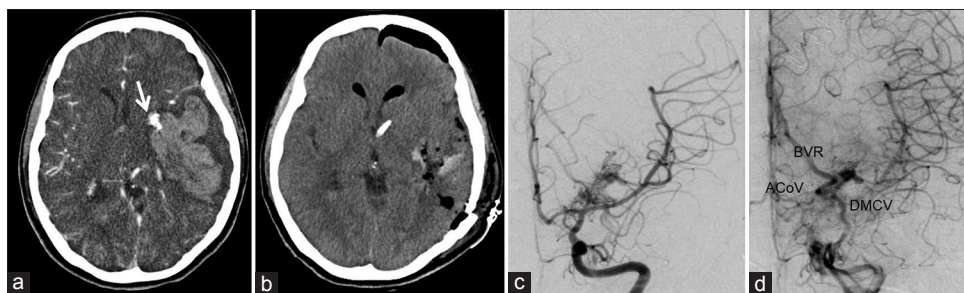


Figure 1: A computed tomographic scan showing an enhanced mass in the anterior perforated substance (a) associated with a huge hematoma in the basal ganglia and temporal lobe. Post operative CT showed the hematoma was completely removal (b). Angiograms revealed a nidus fed by the anterior choroidal arteries (AChAs) and the lateral lenticulostriate arteries (LSAs) (c) and drained by the basal vein of Rosenthal (BVR), the deep middle cerebral vein (DMCV), and the anterior communicating vein in the delayed arterial phase (d)

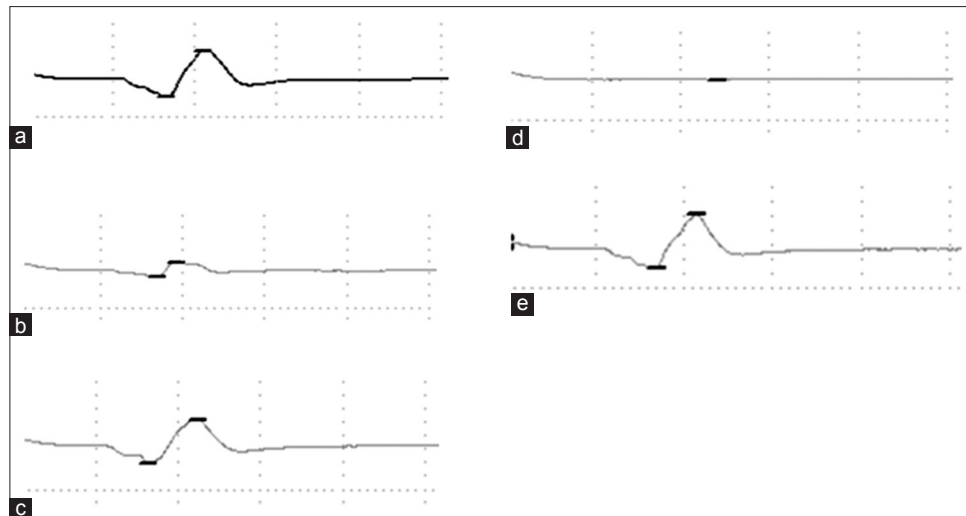


Figure 2: The MEP records showing the change before and after clamping an AChA (a and b) and an LSA (c and d). The amplitude decreased 1 minute after occlusion of one of the four AChAs (b). The amplitude decreased 5 minutes after occlusion of the most lateral LSA (d). They recovered soon after the declamp and did not change at the time of the dural closure (e)

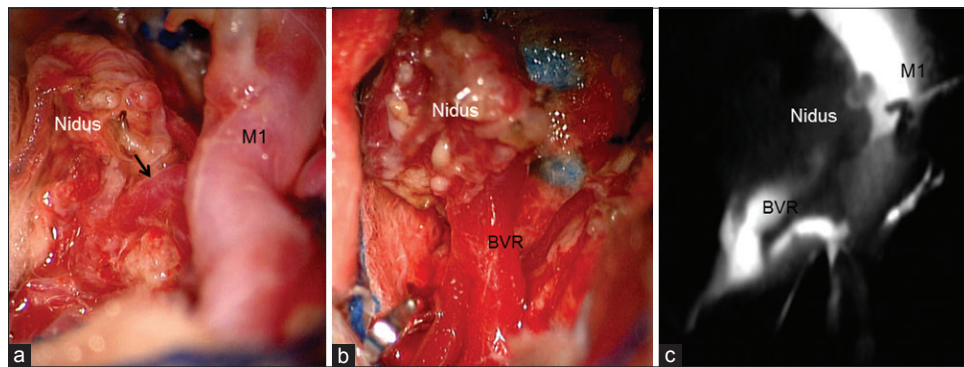


Figure 3: Intraoperative photos. One of the LSAs was found to enter the nidus (arrow) but proved to be a passing artery (a). The nidus was demarcated, and the BVR remained (b). The intraoperative indocyanine green (ICG) videoangiography showed that the LSA remained and reflux of ICG was found in the BVR (c)

green (ICG), and that there was a reflux of the ICG in the BVR [Figure 3c]. The nidus was finally removed after dividing the main drainer. The MEP was maintained at the time of dural closure [Figure 2e]. The fetal heart rate was maintained in the normal range (<80 bpm) throughout the procedure. A postoperative magnetic resonance image revealed small infarcts in the nonfunctional areas, which did not produce any additional symptoms. Pre [Figure 4a] and postoperative [Figure 4b] angiograms showing the AVM had completely disappeared, and two AChAs and one LSA were preserved. The patient successfully delivered a term neonate by cesarean section 122 days after the operation. She could take care of her baby herself, and the child developed normally. The mother, however, suffered dysgraphia due to initial intracerebral hemorrhage that remained 1 year after the onset.

DISCUSSION

AVM has a hemorrhage rate of 8.1% for each pregnancy or 10.8% per year.^[7] The rerupture rate increases during

pregnancy in patients with ruptured AVMs, although pregnancy does not increase the incidence of hemorrhage in unruptured AVMs.^[9] Therefore, a preventive treatment of ruptured AVMs should be considered to save mothers.^[4,7] In the present case, we evacuated a hematoma to prevent a brain herniation, and the AVM was removed 10 days later, whereupon the patient recovered motor and speech functions. If she, her husband, and family would have opted for an abortion, radiotherapy would have been recommended. If the hemiplegia and aphasia were complete, with little hope of recovery, total removal of the nidus would not be difficult because cutting the perforating arteries would not induce further neurological deficits.

It usually takes 2 or 3 years for an AVM to resolve after radiosurgery. Therefore, AVMs are not obliterated during pregnancy. Unless the AVM is completely resolved, the risk of bleeding remains.^[3] The radiation dose in stereotactic radiosurgery for an AVM should be below the safety dose in pregnant women.^[11] Endovascular therapy was reported to be effective as a presurgical strategy

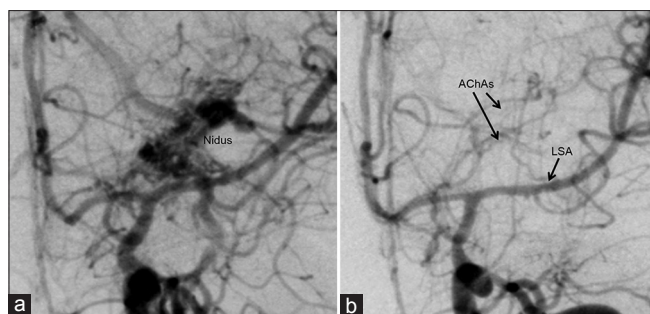


Figure 4: Pre (a) and postoperative (b) angiograms showing the nidus completely obliterated, and two AChAs and a lateral LSA were successfully preserved (arrows)

even in pregnant women.^[1,2] However, the safety of using contrast media remains uncertain during pregnancy.^[16]

Potts *et al.*^[14] reported 48 deeply located AVMs which were classified into three groups based on their locations: the basal ganglia, thalamus, or insula. Among those 48, 10 AVMs located in the basal ganglia were surgically removed. Endovascular treatment was rarely used. We also found it difficult to cannulate a microcatheter into the small feeders. A similar AVM occupying the anterior perforated substance was reported by Garrido *et al.*^[6] in 1978. The AVM was fed by the LSAs and drained into the BVR. However, the nidus was located slightly laterally compared to that in the present case in which there was a medial shift of the LSAs. The nidus was successfully removed without neurological damage.

MEP monitoring proved to be essential to avoid damage to the AChAs and LSAs in the present case. Three perforators in and around the nidus were preserved because the amplitudes disappeared in two AChAs and in a single LSA. Suzuki *et al.*^[15] analyzed the blood flow insufficiency of the AChAs using intraoperative MEP monitoring, which revealed that the MEP disappeared or decreased in 20 patients. One of those 20 patients in whom the MEP disappeared without recovery suffered severe postoperative motor paresis. Four patients whose MEP disappeared temporarily suffered mild hemiparesis transiently.

How long is necessary to confirm the tolerance of the perforating arteries before their permanent occlusion? Horiuchi *et al.*^[8] analyzed 10 cases in which the MEP was changed by the blood flow insufficiency of the middle cerebral arteries. In 7 cases, the amplitude decreased or disappeared within 5 min and in 4 cases within only 1 min.^[8] Therefore, we speculated that we should measure the amplitude twice at 1 and 5 min after the temporary occlusion. The amplitude disappeared when one of the AChAs was clamped for 1 min but did not change at 1 min but disappeared at 5 min in the most-lateral LSA. Those amplitudes recovered soon after the declamp. If we cut the LSA, based only on the MEP finding at 1 min after the clamp, a cerebral infarct would have been induced in the territory of the artery.

During pregnancy, anesthetic gas is generally used to keep the uterus relaxed. However, the anesthetic gas disturbs MEP monitoring due to its muscle relaxing potency. We asked the anesthesiologist to use intravenous anesthesia and to monitor the fetal heart throughout the procedure. Bradycardia with <80 bpm for 2 min indicates fetal hypoxic damage.^[13] In the present case, the fetal heart rate was always kept above 80 bpm; and, fortunately, a spontaneous abortion did not occur. The heartbeats can be detected intraoperatively in a fetus older than 16 weeks of pregnancy, although it is more difficult in younger ones.^[5]

CONCLUSION

We described a rare case of a 31-year-old woman in the 15th week of gestation, presenting with an AVM in the APS who was comatose and hemiplegic. After regaining consciousness and motor function, the AVM was successfully removed with minimal neurological damage; she gave birth to a normal, healthy baby by caesarian delivery. This kind of surgery can be successfully performed provided the nidus is small, MEP is monitored efficiently, and the fetus is older than 15 weeks.

Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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