Ruptured arteriovenous malformation and subarachnoid hemorrhage during emergent cesarean delivery: A case report

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Cerebral arteriovenous malformations (AVMs) are formed from a vascular plexus of direct arterial-venous connections that progressively dilate, making them prone to rupture. They are frequently asymptomatic and often remain undiagnosed until they present with associated symptoms of headaches, seizures, neurological deficits, or hemorrhages. Occurrence of headache during pregnancy and labor is associated with several diverse etiologies, making definitive diagnosis extremely difficult.

This case report describes the anesthetic management of a 31-year-old laboring patient who first complained of headache, then suffered an acute subarachnoid hemorrhage secondary to rupture of a previously undiagnosed AVM during emergent cesarean delivery.

Key words: **Anesthesia, arteriovenous malformation, headache, pregnancy.**

Cerebral arteriovenous malformations (AVMs) are usually formed in the third week of fetal development and reportedly occur at a rate of 1 in every 1,000 to10,000 people.[1,2] They are frequently asymptomatic and therefore remain undiagnosed until they present clinically in young adulthood with headaches, seizures, neurological deficits, or hemorrhages.[3] Intracranial AVMs are formed from a vascular plexus of direct arterial-venous connections that lack an intervening capillary bed.[4-6] These connections result in a high flow, low pressure vascular pathway that cause associated arteries to become progressively dilated.[6] When an occult AVM is discovered during pregnancy, it usually presents as a cerebral hemorrhage.[3,7] The incidence of AVM rupture during pregnancy is 1 in every 10,000 pregnancies.[1] The following is a case report of acute subarachnoid hemorrhage following rupture of a previously undiagnosed AVM that presented during an emergent cesarean delivery.

Case summary

A 31-year-old, gravida 1, para 0, female, 64 inches tall and weighing 180 pounds at 41 weeks' gestation, presented for induction of labor for oligohydraminos. Medical history was significant for migraine headaches first diagnosed at the age of 14. The patient described these headaches as frontal and primarily located in the left temporoparietal region. In addition, the patient described a "blind spot" which was secondary to "a constricted ophthalmic vessel." Her prenatal course was unremarkable with the exception of frequent urinary tract infections and lower extremity edema.

The induction of labor was initiated with cervical application of misoprostil. Preinduction blood pressure was 121/72 mm Hg, and her urine was negative for protein. Complete blood count revealed hemoglobin, 13.3; hematocrit, 38.3; and platelets, 147,000. A spontaneous rupture of membranes occurred at approximately 10:45 PM on the day of admission. By 4:00 AM the next day, the patient's labor was well established and her cervix had dilated to 4 cm. An epidural anesthetic was requested for labor pain. Immediately prior to the epidural placement, the patient's blood pressure was 141/79 mm Hg, and her heart rate was 68 beats per minute. A fluid bolus of 2,000 mL of lactated Ringer's solution was administered, and the patient was placed in the left lateral decubitus position. The epidural catheter was placed midline, in the L3-4 interspace, atraumatically, on the first attempt via a 17-gauge Hustead needle. A test dose of 45 mg of lidocaine with 15 µg of epinephrine was administered. After an appropriate interval for safety evaluation, the epidural catheter was dosed with 8 mL of 0.25% bupivacaine and 20 µg of sufentanil. A sensory level to T10 dermatome was obtained. A continuous infusion was initiated with 0.125% bupivicaine with 0.5 µg of sufentanil per milliliter at 10 mL/hr. The patient remained comfortable (pain score of 1 out of 10) and in active labor for the following 6 hours. Her hemodynamic status was stable with blood pressures approximately 140s/70s.

At approximately 10:40 AM, the patient complained of a sudden onset of severe headache that was first described as occipital and then frontal. She denied scotomata, nuchal pain, photophobia, or a difference in the quality of the headache with positional changes. She reported the headache was not consistent with her previous migraines, and that previous migraines had been resolved with 2 tablets of isometheptene (Midrin). The blood pressure recorded at the time of the complaint was 172/90 mm Hg, and her heart rate was 72 beats per minute. A gross neurological examination revealed that the patient was oriented to person, place, and time. Her pupils were equal and reactive to light. The level of sensory anesthesia remained at T10, and motor ability in all 4 extremities remained equal and strong. Although no signs or symptoms of catheter migration had been observed, the epidural infusion was discontinued, and the catheter was aspirated to rule out migration into the subarachnoid space or an epidural vein. No cerebral spinal fluid or blood was detected. The obstetric staff was consulted and chose to manage the headache pain with 50 µg of fentanyl intravenously. No relief of headache pain was obtained. Because isometheptene had been successful in managing her prior migraines, she was given 2 tablets of this medication for suspected migraine with atypical presentation.

At noon the patient complained of labor pain and a sustained intractable headache. The patient's blood pressure remained elevated at 177/90 mm Hg, and the fetal heart tones had risen from 140s to 170s. The epidural infusion that had been turned off was now restarted with a bolus of 10 mL of 1% lidocaine with 100 µg of fentanyl. Within 10 minutes a T10 sensory level of anesthesia was obtained. Due to her increasing blood pressure and unrelenting severe headache, the obstetric staff initiated a magnesium bolus of 4 g for suspected onset of preeclampsia. This was followed by a magnesium infusion at 2 g/hr. A complete blood count and urinalysis were obtained and sent for immediate analysis to support suspicion of preeclampsia. At 12:25 PM, the obstetric staff requested a cesarean delivery for failure to progress, suspected severe preeclampsia, and suspected chorioamnionitis. The patient's blood pressure had continued to rise (160-180s/70-103), and her temperature was now 38.5°C. She was given an additional 13 mL of 2% lidocaine with 1:200,000 epinephrine in divided doses over 10 minutes. A surgical block to T2 was obtained. A gross neurological examination remained unchanged, and the patient reported her headache was "slightly better." She demonstrated generalized drowsiness, but this was thought to be a typical response to magnesium therapy. The skin incision was made at 12:51 PM, and at 12:56 PM a healthy baby girl was delivered. At 1 minute and 5 minutes, the Apgar score was 9 and 9. A pitocin infusion was initiated to preserve uterine tone.

Despite the pitocin infusion, the surgeons complained of poor uterine tone. Hemabate, 250 µg, was injected into the uterus without any notable improvement in uterine tone. Methergine was not administered because of its potential to further increase blood pressure and induce eclampsia in a preeclamptic patient. At 1:05 PM, the surgeons reported "oozing" on the surgical field. Because coagulapathy is a known complication of preeclampsia, a coagulation profile was drawn and sent for immediate analysis. The patient's blood pressure was now noted to be decreased from 120s/60s to 80s/40s. She responded to intermittent doses of ephedrine in 5-mg increments (total dose, 25 mg) and 50 µg of phenylephrine; her blood pressure stabilized at 90s/40s. At 1:30 PM, the patient became unresponsive to loud verbal stimuli and pain. Her right pupil was dilated to 7 mL and was nonreactive to light. Her left pupil was 3 mL and remained reactive to light. She continued to breathe spontaneously, maintaining oxygen saturation of 100%. There were no electrocardiogram changes noted.

The patient was immediately given 250 mg of sodium pentothal and intubated with a 7.0-mm endotracheal tube to secure her airway. She was then hyperventilated to an end-tidal carbon dioxide of 25 to decrease intracranial pressure. A neurology consultation was requested immediately. At 1:35 PM, a neurologist was present in the operating room and recommended mannitol (1 g/kg) and a computed tomography (CT) examination as soon as the surgical incision was closed. The patient was now demonstrating decorticate vs decerebate posturing. She was given 80 g of mannitol and 25 mg of rocuronium for pharmacological paralysis. Continued hyperventilation to an end-tidal carbon dioxide of 20 to 25 was maintained and at 2:05 PM the patient was transported to the CT scanner.

During transfer from the gurney to the CT table, a large amount of vaginal bleeding was noted, as well as oozing from the surgical site. Previously drawn coagulation results were now available and revealed platelets of 86,000 (normal, 150,000 to 400,000), prothrombin time of 15.4 (normal, 10 to 13 seconds), partial thromboplastin time of 42 (normal, 20 to 34 seconds), and fibrinogin of 74 (normal, 200 to 400 mg/dL). These laboratory results are consistent with disseminated intravascular coagulation, a condition associated with both preeclampsia as well as an intracerebral injury. Two units of crossmatched packed red blood

cells were ordered, and a hematology consultation was requested to assist in managing the coagulapathy.

Computed tomography revealed a midline shift with blood in the right Sylvan fissure, the right temporal lobe, and the right frontal lobe. A neurosurgery consult was requested immediately. At 2:45 PM, the neurosurgeon requested a right carotid angiogram to rule out a preserved middle cerebral artery aneurysm vs a ruptured AVM. The patient was transported to angiography, and a limited angiogram was performed. A middle cerebral artery aneurysm was ruled out. The right pupil remained dilated to 7 mL, and the left pupil was now dilated to 5 mL.

Informed consent was obtained from the patient's husband and at 3:45 PM the patient underwent emergency craniotomy for evacuation of subarachnoid hematoma and resection of AVM and infarcted brain in the right temporal and frontal lobes. Intraoperatively, her coagulapathy was treated with 4 units of fresh frozen plasma, 10 units of cryoprecipitate, a 12 pack of platelets, and 2 units of packed red blood cells. Immediately postoperatively her left pupil size had returned to baseline, but the right pupil remained fixed and dilated. A postcraniotomy CT revealed improvement in intracranial midline shift and complete evacuation of subarachnoid hematoma without evidence of further bleeding. The patient remained intubated in transport to the intensive care unit where she was responsive only to noxious stimuli. By 5:00 AM on postoperative day 1, resolution of her coagulapathy had occurred without administration of additional blood products.

The patient remained in the intensive care unit, revealing gradual improvement in neurological status, for the following 19 days. After regaining consciousness, she was noted to have only limited use of her right upper extremity. Over time she regained considerable use of this extremity; however, she demonstrated very little movement of her right lower extremity, minimal movement of the left upper extremity, and no movement in the left lower extremity. Her speech was limited to only a few single words, but she was able to communicate more extensively by writing. She had difficulty swallowing and required placement of a gastrostomy tube for feedings. The patient was eventually transferred to an inpatient rehabilitation hospital where she is demonstrating slow, continued improvement with aggressive physical therapy.

Discussion

Patients with unrecognized cerebral AVMs are usually asymptomatic and thus at risk for sudden acute hemorrhage with potentially severe or even fatal neurological outcome at any time. Whether that risk is increased in pregnant women is a source of controversy among investigators.[3,7] It is hypothesized that the increase in cardiac output associated with pregnancy, and especially labor, will stress a fragile AVM and render it more susceptible to rupture.[5,7,8] One investigator reports pregnant women have a 4 times' greater risk of AVM rupture than nonpregnant women.[7] However, another disputes this long accepted association and found that pregnancy did not contribute to increased risk of hemorrhage from AVM rupture.[3]

This case report demonstrates a differential diagnosis fraught with uncertainty. Occurrence of headache is associated with several diverse etiologies, making definitive diagnosis extremely difficult (Table).[4,9] Pregnancy adds even more variables to consider. Our patient had a history of migraine headaches. Migraine headaches are easily triggered by emotional or physical stress, lack of sleep, and missed meals, all common to the laboring patient.[4] Although her headache pain was not consistent with previous migraines, an attempt to exclude migraine as an etiology was made with administration of isometheptene, a drug she had used successfully in the past. Headaches also are associated with dehydration. Our patient received several fluid boluses during placement and maintenance of regional anesthesia. Hypertension is a known cause of headache. Our patient's hypertension occurred in conjunction with the onset of her headache, leading the obstetrical staff to suspect and treat her for preeclampsia. Magnesium bolus and infusion was initiated. Common side effects of magnesium include hypotension, weakness, and sedation, possibly obscuring her neurological examination.[9] Dural punctures are a known potential risk of regional anesthesia that can cause sudden or delayed headache[9]; however, dural puncture did not occur during placement of the epidural catheter. Catheter migration into the subarachnoid space also was ruled out.

The character of our patient's headache was sudden and severe, first occipital and then bilateral frontal. Migraine headaches are lateralized or generalized and are usually dull or throbbing.[4] Postdural puncture headaches and those associated with preeclampsia are also dull or throbbing.[9] Headaches associated with AVM are nonspecific in character and can mimic migraines.[4] Subarachnoid hemorrhage, however, is characterized clinically by sudden onset of a headache with unusual severity, often described as explosive and unlike any previous headaches experienced.[4] This headache is commonly followed by nausea and vomiting, then impairment of consciousness that is transient or progresses to coma and then death.[4]

Table. Differential diagnosis of headache[4,9]

	Etiology	Signs and symptoms
Tension headache	Stress, fatigue, noise, glare, hunger	Generalized pain, vague, nonspecific, vice-like, or "tight"; most intense at neck or occiput; no focal neurologic symptoms
Migraine headache	Dilation and excessive pulsation of cerebral vasculature	Gradual onset, lateralized or generalized pain, throbbing, episodic; associated with anorexia, nausea and vomiting, photophobia, blurred vision, aura, family history of migraines
Postdural puncture headache	Leakage of cerebral spinal fluid through dural puncture	Generalized pain, dull, throbbing, aggravated by erect posture, alleviated by recumbency
Preeclampsia headache	Generalized vasospasm and cerebral edema	Generalized pain, dull, throbbing; accompanied by hypertension, hyperreflexia, scotomas, proteinuria, and generalized edema
Subarachnoid hemorrhage headache	Usually results from rupture of aneurysm or arteriovenous malformation	Sudden onset, severe pain, meningeal irritation, impairment of consciousness, focal or generalized seizures possible

The increased blood pressure experienced by our patient in conjunction with her onset of headache was most likely related to a physiological response to improve cerebral perfusion pressure in the presence of increased intracranial pressure.[10] When an AVM ruptures, as in this case, the acute high pressure leakage of blood into the subarachnoid space markedly increases the intracranial pressure.[9] This leads to early symptoms of headache and sedation.[9] These were, in fact, the presenting symptoms experienced by our patient. The increased intracranial pressure also leads to severe intracranial tissue damage, a known cause of disseminated intravascular coagulation.[11] This was first evidenced by us as "oozing" on the surgical field during the cesarean delivery. This observation was rapidly followed by a profound change in neurological status, indicated by pupil changes and a decreased level of consciousness. Such symptoms require immediate and aggressive investigation and treatment.

Because of the similarities in differential diagnosis with headaches related to normal or complicated pregnancy, delay in correct diagnosis and management can easily occur. If subarachnoid hemorrhage is not immediately treated with decompression, rapid brain displacement will result in death.[9] For this reason, early neurological consultation should be considered whenever the etiology of a headache is uncertain.

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