Ochsner Journal 16:542–544, 2016

Hemorrhagic Stroke Resulting From Venous Malformation at 20 Weeks of Pregnancy

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Background: Stroke in the pregnant patient is not a common occurrence. Despite its relative rarity, stroke during pregnancy is associated with high morbidity and mortality for both mother and infant. We report the case of a patient who experienced a hemorrhagic stroke during pregnancy because of venous cavernoma.

Case Report: A 34-year-old patient, gravida 5 para 1-0-3-1, presented to labor and delivery triage at 21 weeks, 0 days' gestation with the concern of sudden-onset right-sided facial, arm, and leg numbness and weakness. Intracranial imaging via magnetic resonance imaging demonstrated a small left midbrain venous cavernoma in the periaqueductal region of the posterior lateral cerebral peduncle area with evidence of edema and focal hemorrhage. During a 3-day hospital admission, the patient's deficits slowly improved, and she was discharged home. She subsequently delivered vaginally without incident. Six days postpartum, the patient presented with symptoms that were similar to her initial presentation. She was correctly diagnosed with migraine with aura after the appropriate neuroimaging studies did not show an acute stroke.

Conclusion: This case demonstrates the similarities between the perilous diagnosis of stroke and the more routine diagnosis of migraine. The case also highlights the need for caution and for a multidisciplinary treatment approach when the diagnosis of stroke is considered, particularly in the pregnant patient.

Keywords: *Hemangioma–cavernous, intracranial hemorrhages, pregnancy, stroke, vascular malformation*

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INTRODUCTION

Stroke in the pregnant patient is not a common occurrence. The incidence is somewhat debated, but recent evaluations suggest the incidence is from 4-7[1] to 25-34[2] strokes per 100,000 deliveries. Despite its relative rarity, stroke in the pregnant patient is associated with high morbidity and mortality for both mother and infant.[1,3,4] The adverse outcomes for the infant seem to be mediated in part by increased preterm birth, higher rate of cesarean delivery, and increased frequency of neonatal intensive care unit admission.[3] Both ischemic and hemorrhagic strokes can occur in pregnancy, although hemorrhagic stroke can be particularly dangerous for the pregnant patient. Hemorrhagic stroke has been documented to occur in the range of 5-35 per 100,000 deliveries[1,3-5] with maternal and fetal mortality rates as high as 63% and 27%, respectively.[1] We report the case of a patient who experienced a hemorrhagic stroke during pregnancy because of a venous cavernoma.

CASE REPORT

A 34-year-old patient, gravida 5 para 1-0-3-1, presented to labor and delivery triage at 21 weeks, 0 days' gestation with the concern of sudden-onset right-sided facial, arm, and leg numbness and weakness. The weakness was associated with an intractable headache, photophobia, phonophobia, nausea, and vomiting. Ambulation and speech were unaffected. She denied any trauma or inciting event but reported a recent hiking trip with a 5-hour car ride. Her medical history was significant for migraine headaches, Hashimoto disease, osteopenia, ulcerative colitis, and secondary infertility. Her obstetric history was significant for 2 early pregnancy losses followed by a normal spontaneous vaginal delivery at term 4 years prior. After that successful pregnancy, she developed ulcerative colitis 10 days postpartum and was diagnosed with new-onset Hashimoto disease at 6 months postpartum. During the course of her treatment, she developed and was treated for osteopenia. She was subsequently diagnosed with secondary infertility related to hypothalamic amenorrhea after she was unable to spontaneously conceive for 1 year. She was treated for infertility and successfully conceived via in vitro fertilization. However, she had a first trimester miscarriage on her first round of therapy that required suction dilation and curettage. She then conceived the current pregnancy via ovulation induction with intrauterine insemination. Her current medications included levothyrox-

ine 112 mcg by mouth daily, mesalamine 1.2 g by mouth daily, and a prenatal vitamin. She reported allergies to ibuprofen, hydromorphone, and nitrofurantoin.

Significant family history included her mother with hypothyroidism and deep vein thrombosis while on estrogen, her sister with hyperthyroidism, and her grandmother with possible aneurysm or stroke. The patient had never been tested for inherited thrombophilias prior to presentation. Her social history was significant for aggressive daily exercise; the patient reported running 5-7 miles daily. She denied tobacco or illicit drug use. Her surgical history was significant for the aforementioned suction dilation and curettage.

On physical examination, the patient's vital signs were within normal limits, and fetal heart tones were verified at 150 beats per minute. No erythema, swelling, or tenderness was seen in the lower extremities. Neurologic examination was positive for slight right-sided ptosis, numbness of the right face, right-sided pronator drift, and strength 4/5 on the right side and 5/5 on the left side. Her ambulation was intact with minor balance issues. Physical examination was otherwise noncontributory.

Intracranial imaging via magnetic resonance imaging (MRI) demonstrated a small left midbrain venous cavernoma in the periaqueductal region of the posterior lateral cerebral peduncle area with evidence of edema and focal hemorrhage. Neurosurgery was consulted, and the patient was immediately transferred to the neurocritical care service for continued workup and treatment. The patient was admitted and observed for 3 days. Her deficits slowly improved, and she was discharged on hospital day 3. She was followed by neurology during the remainder of her pregnancy, and no contraindication to a vaginal delivery was determined. She was subsequently admitted in active labor at 38 weeks, 5 days' gestation. When she reached the second stage of labor, she pushed for 23 minutes to deliver a 2,910-g infant with Apgar scores of 9 and 9 at 1 and 5 minutes, respectively. Her postpartum course was uncomplicated, and she was discharged home on postpartum day 2.

On postpartum day 6, she presented to the emergency department with left eye visual disturbances (decreased acuity with floaters), headache, and nausea. Head computed tomography was negative for hemorrhage. MRI showed a more obvious hemorrhage from the cavernous malformation compared to prior imaging, possibly representing interval bleeding. However, the hemorrhage was not considered to be clinically significant at the time, and the patient was diagnosed with migraine with aura. A follow-up MRI 1 month later demonstrated shrinking of the lesion consistent with resorption of old blood. The patient continues to follow up with the neurology and neurosurgery services and has been advised that definitive surgical treatment is not warranted unless the lesion begins to clearly enlarge or symptoms significantly worsen.

DISCUSSION

Although stroke is relatively uncommon during pregnancy, the physiologic changes that occur with gestation, such as a high estrogen state and the growing uterus and fetus, theoretically increase the risk of stroke. The vessel wall remodeling that occurs during pregnancy makes vessels vulnerable to the increasing hemodynamic stresses that occur as the pregnancy progresses and delivery occurs.[1] Interestingly, the postpartum period is particularly dangerous because most pregnancy-related strokes, both ischemic and hemorrhagic, occur during this time.[1,5] Compared to age- and race-matched nonpregnant controls, the relative risk for ischemic and hemorrhagic stroke is reported as high as 8.7 (95% confidence interval [CI], 4.6-16.7) and 28.3 (95% CI, 13-61), respectively, in the postpartum period.[5]

Stroke during pregnancy has been related to chronic hypertensive disorder, hypertensive disorders of pregnancy, gestational diabetes, increasing maternal age, smoking, thrombophilias, and migraines, among other conditions.[3,4,6] Although the numbers in the literature vary, ischemic strokes, including cerebral venous thrombosis, seem to be more common than hemorrhagic strokes during pregnancy.[1,4,5] Hemorrhagic strokes have been reported to comprise 25% of strokes occurring at any time during pregnancy[4] and 40% of antenatal strokes.[3] Another US study based on the Nationwide Inpatient Sample database found 15.7% of antenatal, 10.8% of delivery-related, and 35.6% of postpartum strokes to be hemorrhagic in nature.[2] Ischemic strokes are most commonly related to preeclampsia/eclampsia, venous sinus thrombosis, and cardioembolism.[1,3] Hemorrhagic strokes are often related to preeclampsia/eclampsia as well. In addition, several types of vascular malformations can cause hemorrhagic strokes in both pregnant and nonpregnant patients, including cerebral aneurysm, arteriovenous malformation (AVM), and cavernous malformation.[1,7,8]

Management of hemorrhagic stroke during pregnancy differs based on the etiology of the stroke, but consultation with the neurology and neurosurgery teams is essential when developing a treatment plan. In some cases, such as a patient with a newly discovered aneurysm, early surgical intervention significantly decreases maternal and fetal mortality.[1] With hemorrhages caused by other forms of vascular malformation, the need for surgical intervention may not be as urgent, depending on size, location, and symptoms.[1,9] AVM and cavernous malformation have detection rates of 1.1 and 0.6 per 100,000 adults per year, respectively.[7] Evidence suggests that when an AVM bleeds in a pregnant patient, the risk of a subsequent bleed from the same lesion may be higher compared to an isolated hemorrhage in a nonpregnant patient with an AVM.[1] Individuals with cavernous malformation can have multiple lesions, a condition that is typically associated with familial inheritance, or a single lesion, which is usually sporadic.[7] The risk of bleeding from a known cavernous malformation does not appear to be increased during pregnancy.[8,9] However, when familial and sporadic lesions were analyzed separately, familial lesions had a slightly higher risk of hemorrhage at 3.6% per pregnancy compared to 1.8% for sporadic lesions.[8] In the 2013 report by Kalani and Zabramski of cavernous malformations during pregnancy, 149 of 168 deliveries occurred vaginally, with no delivery-associated cerebral hemorrhages.[8] Overall, the data seem to suggest that vaginal delivery should not be contraindicated for patients who experience hemorrhagic stroke and have stabilized by the time delivery is imminent.[1,8,9] However, if an urgent obstetric issue arises prior to neurosurgical intervention or during an acute bleed, delivery

via cesarean section may be recommended, followed immediately by surgical control.[1] Whether or not acute hemorrhage is present, neurosurgical evaluation for individualized risk is always recommended when a cavernous malformation or any vascular malformation is discovered during pregnancy.[9]

Interesting to note is the patient's history of autoimmune disorder, as she had acute onsets of Hashimoto disease and ulcerative colitis after her first delivery. While no link has been established between cerebral cavernous malformation and either Hashimoto disease or ulcerative colitis, case reports have suggested a link between ulcerative colitis and gastrointestinal-related vascular malformations such as cavernous hemangiomas.[10,11] The authors of one these reports suggest that the inflammation in ulcerative colitis may interfere with vascular integrity, allowing cavernomas to form in the gastrointestinal tract.[11]

In our case, the patient had only one lesion visualized, and it was thought to be sporadic. After careful neurologic and neurosurgical evaluation, her risk of recurrent hemorrhage during pregnancy was estimated to be quite low, and she delivered vaginally without incident. However, she presented during the high-risk postpartum period with symptoms that were similar to her initial presentation. She was correctly diagnosed with migraine with aura after the appropriate neuroimaging studies did not show an acute stroke.

CONCLUSION

This case demonstrates the similarities between the perilous diagnosis of stroke and the more routine diagnosis of migraine. The case also highlights the need for caution when the diagnosis of stroke is considered, particularly in the pregnant patient. When dealing with complex diagnoses in pregnant patients, a multidisciplinary approach that includes all relevant subspecialties is indicated to achieve the best outcome.

ACKNOWLEDGMENTS

This work was presented and discussed at the Obstetrics and Gynecology Morbidity and Mortality conference, sponsored by the Ochsner Obstetrics and Gynecology Department, held at Ochsner Baptist Medical Center on February 11, 2015.

The authors have no financial or proprietary interest in the subject matter of this article.

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