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Case Report

Postpartum Syncope with a New Onset of Headache: a Case Report

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Postpartum period is an especially vulnerable period of time. Headache, dizziness, and loss of consciousness are not uncommon in patients who are immediately postpartum. In order to rule out potentially fatal causes such as intracranial hemorrhage, monitoring of clinical signs and offering diagnostic studies are of essential. Here we present a case with a new headache in the postpartum period after a syncope fall accident, and an intracranial vascular malformation noted in subsequent radiologic evaluation. A 36-year-old woman without underlying condition developed severe headache following a syncope fall accident after a vaginal delivery. An intracranial vascular malformation was noted in the brain CT scan, and MR imaging and digital subtraction angiography revealed a mixed lesion of a cavernous malformation and a developing venous anomaly. Due to hemodynamic fluctuation, headache or syncope is not an uncommon sign in the postpartum period. When companied with worrisome features, these symptoms require further investigation. Our patient was diagnosed to have a mixed vascular malformation, which is a coexistence of a developing venous anomaly and a cavernous malformation. After the diagnosis was confirmed, the possible need for surgical intervention and future rebleeding risk, especially in the next pregnancy, should be evaluated. Previous studies concluded that pregnancy, delivery and puerperium were not associated with an increased hemorrhage risk, and the selection of delivery mode in patients with known cavernous malformation should be based on general obstetric indications. Primary care providers need to identify worrisome signs related to intracranial hemorrhage in postpartum patients with syncope, dizziness, or headache. In patients diagnosed to have an intracranial vascular malformation in this period of time, it is most important to make prompt diagnosis and refer her to a specialist for further treatment and counseling.

Keywords: Postpartum period, pregnancy.

1. Introduction

Postpartum period is an especially vulnerable period of time. Headache, dizziness, and loss of consciousness (LOC) are not uncommon complaints in patients who are immediately postpartum. In order to rule out potentially fatal causes such as intracranial hemorrhage, monitoring of patient's vital signs and the development of red flag signs are of essential importance.(1)

Here we present a case with a new postpartum headache after a syncope fall accident, and an intracranial vascular malformation noted in subsequent radiologic evaluation.

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2. Case presentation

Our patient was a 36-year-old woman with no known medical illness. She had one previous uneventful pregnancy with uncomplicated vaginal delivery.

She had been under antenatal care at our hospital, and she did not develop gestational hypertension or proteinuria during antenatal period. Her last antenatal visit was at 37 weeks of gestation. The obstetric sonography showed a normally grown fetus with estimated body weight of 2750 grams in vertex presentation.

She presented to our delivery room at 38+2/7 weeks of pregnancy, with the chief complaint of regular uterine contraction pain and small amount of bloody show. Her physical examination and routine laboratory lab data were all unremarkable. On per vaginal exam, she was found to have a 3-cm dilated, 30% effaced cervix and intact membranes, and the fetal station was -2. Her cervical dilatation was augmented with Hyoscine-N-Butylbromide (Buscopan) intramuscular injection(2). Five hours later, she had an uneventful vaginal delivery of a healthy male newborn weighing 3050 grams. The newborn's Apgar scores were 9 and 10 at 1 and 5 minutes, respectively.

At dawn on her third postpartum day, she experienced LOC on the way back to bed after urinated in the bathroom.

She could recall syncopal prodrome of dizziness as well as visual disturbance (seemingly uneven and darkened floor), but she did not have palpitation, chest pain or cold sweating. She found herself lying on the floor after returning of consciousness. Because there was no witness to the event, the duration of LOC was unknown. Immediately after the event, there was no headache or weakness, and she only complained of lower back pain.

Right after the fall accident, her blood pressure was 117/57 mmHg, heart rate was 52 beats per minute (BPM). On physical examination, she was conscious and oriented. Physical examination showed no laceration or ecchymosis on the scalp, and the neurologic examination was unremarkable. Her lab data revealed that hemoglobin was 11.3 g/dL, white blood cell count was 12.5×10^9/L (Segment 78.2 %, Lymphocyte 16.4 %). The X-ray films, including posteroanterior view of pelvis, lateral view of skull and right hip, showed normal findings.

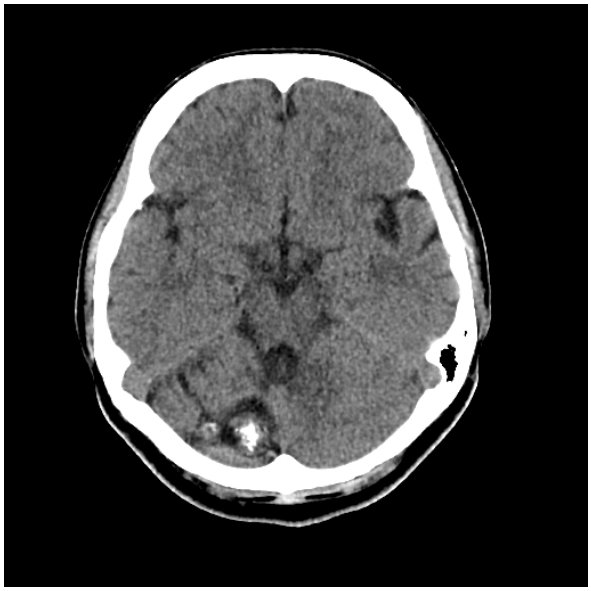


Fig. 1 The CT scan showed a focal abnormal change measuring 2.8 x 2.1cm with a region of calcification surrounded by some lower densities in the right cerebellum, and the differential diagnosis included intracranial tumor and vascular lesion.

Seventeen hours later, in late evening on her 4th postpartum day, her blood pressure was elevated up to 150/73 mmHg. She also had bradycardia with heart rate of 53 bpm. After a few hours, she experienced severe headache and chest tightness along with persistent elevated blood pressure. (BP: 158/80mmHg; Pulse: 62 BPM) Cardiac enzymes were drawn, and were all within normal limits. Due to persisted hypertension, she was given oral antihypertensive agent (Amlodipine 5mg/tab, 1 tablet), and headache and shortness of breath were controlled with acetaminophen and oxygen given through nasal cannula. Four hours after she received Amlodipine, her blood pressure dropped to 93/43mmHg, and her pulse was 64 BPM.

We consulted the neurologists, and arranged non-contrast brain computerized tomography (CT) scan immediately. The CT scan revealed a 2.8 x 2.1cm lesion with surrounding lower densities and some calcifications in the right cerebellum. (Fig.1)

Due to the possibility of intracranial tumor or vascular lesion, the radiologists recommended further evaluation with contrast-enhanced brain magnetic resonance imaging (MRI).

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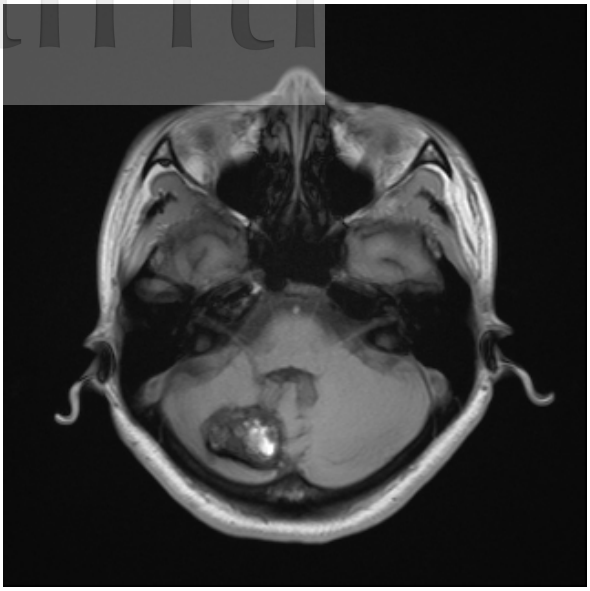


Fig. 2 In this TIWI in MRI, there is bright blood clot accumulation in the cerebellar lesion along with hemosiderin deposition.

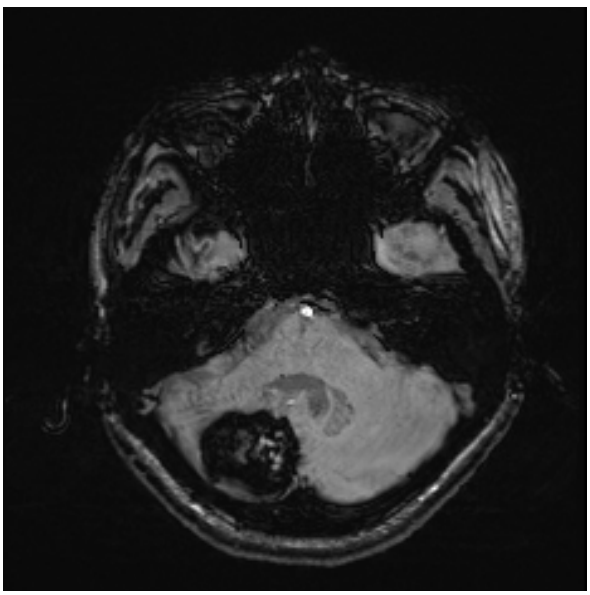


Fig. 3 In this susceptibility-weighted image (SWI), there is a focus of signal loss which is consistent with the region of prior hemorrhage.

The T1 weighted image (TIWI) in MRI showed an intracranial vascular malformation in the right cerebellum with perilesional hemosiderin deposition, associated with previous recurrent hemorrhage. A tortuous and dilated vessel traversing from the right cerebellum to the right transverse sinus was identified. (Fig.2, 3, 4)

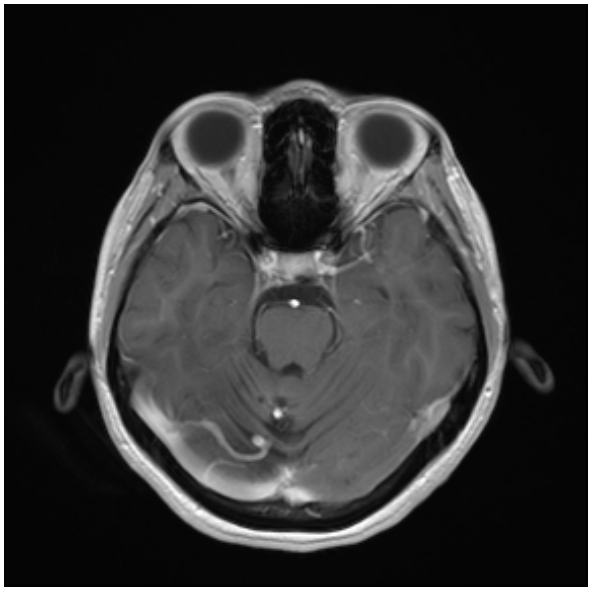


Fig. 4 In this contrast-enhanced MRI image, there is a tortuous and dilated vessel traversing from right cerebellum to right transverse sinus. This is the draining vein of this lesion.

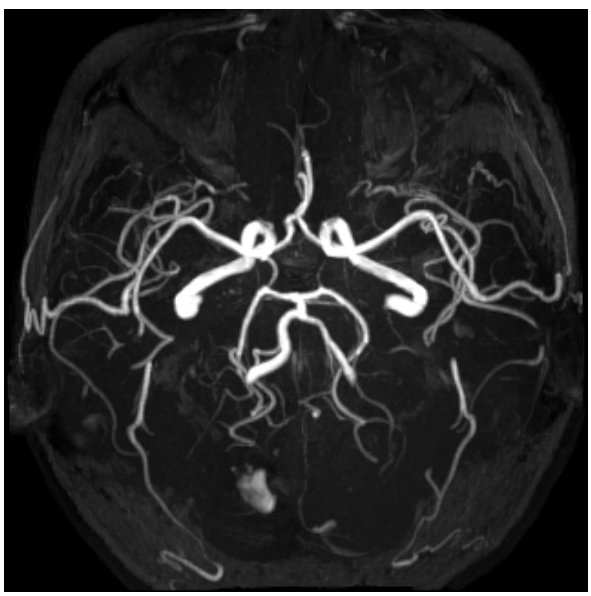


Fig. 5 In this recombination MRA image, we can identify this intracranial vascular malformation in the right cerebellum. There is no evidence of cerebrovascular constriction in this image. A fetal type right posterior cerebral artery, which is a common variant, was noted; the other main trunks of intracranial arteries are preserved.

The differential diagnosis of this intracranial vascular malformation included arteriovenous malformation (AVM) and dural arteriovenous fistula.

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Fig. 6 DSA shows the frontal view of venous phase via right internal carotid injection. Significant engorgement of a vessel in the right cerebellar region was noted, suggesting DVA.

The patient's blood pressure remained within the normal limits after the MRI, and she was discharged on the 5th postpartum day.

She had digital subtracted angiography (DSA) 2 months later. In the venous phase, there was a significantly engorged in the right cerebellar region, and the final diagnosis was developmental venous anomaly (DVA). (See Fig.6)

3. Discussion:

Due to hemodynamic fluctuation, postpartum period is perceived to be a vulnerable period of time. In postpartum patients, syncope is not an uncommon problem as well as a headache and dizziness.

Considering the precipitating factor, the cause of the patient's postpartum syncope can be a neurally mediated syncope, provoked by micturition. Because intrapartum blood loss and postpartum diuresis can predispose the patient to orthostatic hypotension, this could be a possible cause. Although the patient denied history of structural heart disease and arrhythmia, we should still consider the possibility of cardiac syncope. (See Table 1)

Though our patient did not have unstable vital signs, focal neurologic signs or symptoms, in reality,

Table 1. Differential diagnosis of our patient's syncope

Underlying etiology of Syncope	Predisposing factors	Associated symptoms and signs
Neurally mediated syncope	Micturition	Prodrome of visual disturbance (blackout or dark shadow in visual field)
Orthostatic hypotension	Intrapartum blood loss and postpartum diuresis	Onset of symptom after sudden change of position from lying/ sitting to standing
Heart disease	Preexisting structural heart disease, arrhythmia, or peripartum cardiomyopathy	Could be accompanied by chest pain, prodrome of cold sweating and palpitation

Table 2. Worrisome headache features which requires further investigation[4]

Headache with new neurological symptoms
Any previously-diagnosed headache type with change in character or pattern
New headache, especially if fixed to one side
Sudden uncontrollable vomiting
Headache with Change in mental status
Headache not relieved by pain medications
Other concerning medical condition (bleeding disorder, immunocompromised)

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Table 3. Common causes of postpartum headache; MRA= magnetic resonance angiography; MRI= magnetic resonance imaging; MRV= magnetic resonance venography;

Causes of postpartum headache		Associated condition and symptoms/ signs	Further diagnostic survey
Primary headache disorders, such as tension headache, migraine, and cluster headache	Tension headache	Bilateral location; pressing/tightening quality; mild or moderate intensity; not aggravated by routine physical activity such as walking or climbing stairs	Rule out secondary causes of headache
	Migraine	Unilateral location; Pulsating quality; Moderate or severe intensity; Aggravation by or causing avoidance of routine physical activity (e.g., walking or climbing stairs) Accompanied by nausea, vomiting, photophobia or phonophobia	
Postpartum dural puncture headache		Administration epidural anesthesia; "postural effect" in 80% of patients: headache is usually mild or absent when the patient is lying down, but worsens with upright posture	(If empirical treatment fails) MRI with Gadolinium may show meningeal enhancement; lumbar puncture may show low pressure; CT myelogram may show leak; radionuclide cisternogram may pick up slow leaks
Pre-eclampsia		Hypertension, proteinuria or seizure	Assess and monitor for proteinuria, hypertension and hyperreflexia
Vascular etiology	Ischemic stroke	Headache come on abruptly	Brain CT and lumbar puncture to assess for hemorrhage, if negative then MRA and MRV looking for cerebral venous thrombosis
	Hemorrhagic stroke		
	Subarachnoid hemorrhage		
	Vascular malformation (aneurysm, AVM)		
	Hypertension		
	Venous sinus thrombosis		
	Dissection		
	Vasculidities		
Reversible cerebral vasoconstrictive syndromes		Any vasoconstrictive medications	MRI/MRV looking for typical bead-like appearance of vessels seen in vasoconstrictive syndromes
Drug use		Non-prescribed licit and illicit drugs should be considered, e.g. cocaine, pseudoephredrine	Not applicable
Other causes, such as Benign intracranial hypertension, Tumor, and Sinusitis		Not applicable	Not applicable

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Table 4. Differential diagnoses and their characteristics of the patient's intracranial vascular malformation based on the MRA imaging. (6, 8) [13] [14] [15] [16, 17]

Types of intracranial vascular malformation (CVMs)	Incidence	Annual rate of hemorrhage	Pathology and characteristics in imaging
Cavernous malformations	0.5% of the general population	Low; 0.6-1.1% per year	A cluster of closely-packed thin-walled, enlarged capillary channels (caverns) with a single layer of endothelium without mature vessel wall elements or normal intervening brain parenchyma between the interstices of the lesion; Usually present with surrounding hemosiderin due to repeated minor haemorrhages. Lesions are unseen on DSA, thus are called "occult AVM."
Developing venous malformation	>2%	< 0.7% (more in pregnant patients or those having lesions in the posterior fossa)	Clusters of dilated cetripetally draining medullary veins that converge into a common collecting vein, which drains the normal brain parenchyma.
Arteriovenous malformation	10 to 18 per 100000 adults (asymptomatic or symptomatic)	2% to 4%	An abnormal tangle of blood vessels connecting an artery and a vein; present of normal brain tissue between the interstices of the lesion.
Dural arteriovenous fistula	Rare; only account for 10–15% in all CVMs	19.2%	Direct connection of an artery and a vein with no nidus. Dilated vessels (veins) of the cortex, meninges or extracranial tissues are more easily identified on MRI.

a patient with a head injury of undoubted minor mechanism still can rarely develop an intracranial hematoma, deteriorate, and expire. Though difficult to work out the actual effect, concerns about malpractice litigation might have contributed to the rise in using brain CT scan as a means of evaluation for minor head injuries. In our case, we arranged brain CT scan because the patient developed this intractable headache, which is among the worrisome features of postpartum headache (See table 2), in order to rule out intracranial hematoma such as epidural hematoma or subdural hematoma.(3)

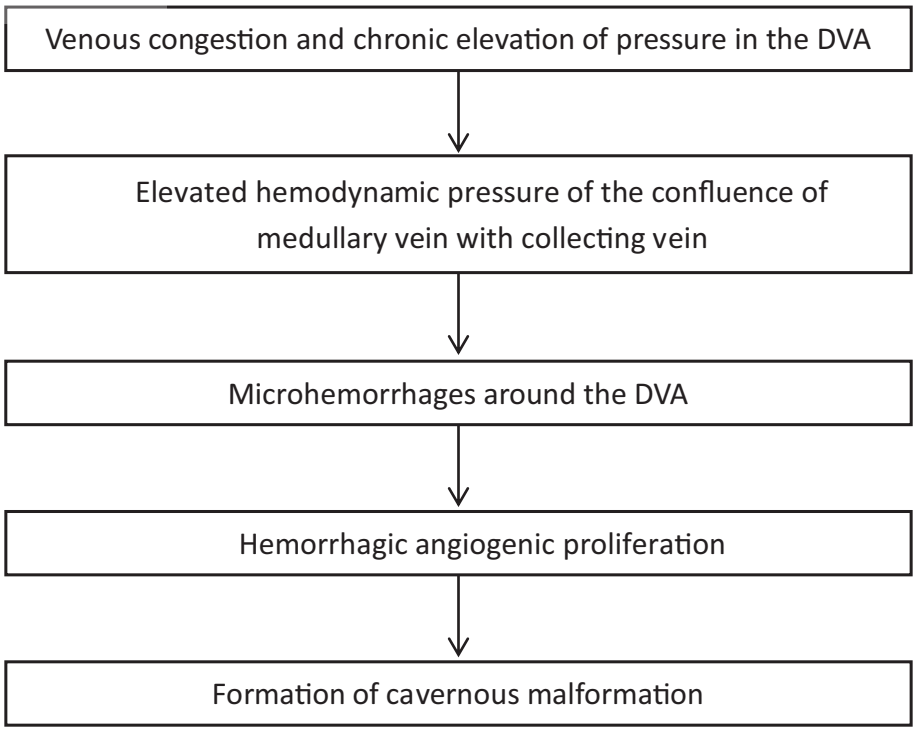
Common causes of postpartum headache include primary headache disorders (such as tension headache, migrane, etc.), postpartum dural puncture headache, pre-eclampsia, intracranial pathology, cerebral venous thrombosis, and the often underdiagnosed reversible cerebral vasoconstriction syndrome. (See table 3)

Our patient denied any other similar episodes of headache in the prenatal period or earlier in her life, but first onset of a primary headache could still be possible. Because she had epidural catheter insertion for painless labor, the diagnosis of postpartum dural puncture headache could not be ruled out. Reversible cerebral vasoconstriction syndrome can develop in the postpartum period, and can also be potentially triggered by factors such as vasoactive medications, including methergine and ergonomine, commonly administered to postpartum patients. However, the brain MRI did not reveal evidence of cerebral vasoconstriction of the main arteries, and thus this differential diagnosis could be ruled out.

The bleeding in this intracranial vascular malformation could be induced by hypertension or minor head trauma (e.g. fall accident), and subsequently cause severe headache because of blood

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Table 5. Hypothesized pathogenesis of a mixed vascular malformation. DVA, developmental venous anomaly



extravasation as a chemical irritant and a secondary inflammatory response in the brain tissue.

Although our patient did not have hypertension or proteinuria during her pregnancy, gestational hypertension and preeclampsia can develop de novo in the postpartum period. Medications which can potentially cause elevated blood pressure, including nonsteroidal anti-inflammatory medications (diclofenac) and ergot alkaloids (methylergonovine) should be discontinued. (4).

On the other hand, although the patient denied any discomforts except for dizziness in the prenatal period, her brain CT and MRI revealed perilesional hemosiderin precipitation and atrophy in the right lobe of cerebellum with exaggerated sulci, which could be the result of chronic recurrent microhemorrhage, hypertension could also have been the result, instead of the cause, of preexisting headache induced by intracranial hemorrhage. The headache developed at the onset of intracranial hemorrhage is associated with chemical irritation by the blood and release of proinflammatory cytokines even in the absence of parenchyma edema with resultant mass effect(5).

Even though our patient's brain lesion is located in the right cerebellum, her cerebellar signs (finger-nose-finger test; heel-to-shin test; Romberg sign) were all negative when we approached her during the hospitalization despite she ever experienced sense of imbalance earlier in pregnancy. In order to clarify the relationship of patient's sense of instability and the cerebellar lesion, we could arrange electrocephalogram during the headache attack.

Based on the MRI studies, the differential diagnoses of her intracranial vascular malformation are: cavernous malformations with an occult AVM, AVM, and arteriovenous fistula. (See table 4).

In the contrast-enhanced magnetic resonance angiography (MRA) image, a dilated vessel traversing to right transverse sinus from right cerebellum which was later proved to be a DVA with DSA. It was seen in the venous phase, unlike an AVM, which would be a dilated vascular lesion seen in the arterial phase. The cavernous malformation, due to its low pressure and slow flow of blood within the lesion, is angiography-occult (not seen in DSA)(6).

DVAs are an extreme anatomical variant of

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medullary veins which drain the normal brain tissue as a compensatory venous drainage system. Although isolated developmental venous anomalies without cavernous malformation might be vulnerable to hemorrhage in hemodynamic stress, it is hypothesized that venous congestion in this condition may induce the formation of cavernous malformation, which is the cause of hemorrhage in a mixed vascular malformation. (Table 5)(6, 7)

About 20% of cases of DSA are associated with cavernous malformations, and are referred to as mixed vascular malformations(8). A recent cohort study concluded that associated DVAs do not correlate with higher hemorrhagic risk or more severe bleedings of cavernous malformation(9, 10).

When counseling patients with a known intracranial vascular malformation who is planning the next pregnancy, we offer statistics regarding to annually bleeding risks, indications of treatment, therapeutic options and delivery routes. Though some literature suggest that there is an increased possibility for a intracranial vascular malformation to enlarge in pregnancy due to the influence of female hormonal fluctuation, studies failed to demonstrate receptors for estrogen or progesterone. Statically, previous studies concluded that pregnancy, delivery and puerperium were not associated with an increased hemorrhage risk in patients with cavernous malformation (11). Before the patient's next planned pregnancy, we should offer surgical consultation if there are indications for definitive treatment, and they include overt intracranial hemorrhage, presence of focal neurologic symptoms, and epilepsy which is intractable under anti-epileptic drugs. Treatment options of definitive treatment for intracranial vascular malformation are surgical resection via craniotomy, embolization via endovascular angiography, and stereotactic radiosurgery. Notably, only the cavernous malformation is resection while the DVA is left untouched due to the risk of thrombosis and infarction in patients with mixed lesions undergoing operation(6).

On the contrary, those asymptomatic intracranial vascular malformations could be managed with conservative treatment and watchful surveillance. If the patient chooses to undergo conservative treatment, MRI within one year prior to the estimated date of confinement is recommended to determine baseline condition of the lesion$_{(12)}$; and MRI can still be the modality of choice to follow her brain lesion in her next pregnancy if she develops new neurologic symptoms, though some experts discourage the use of gadolinium contrast in pregnancy.

Although many obstetricians choose Cesarean sections for patients with intracranial vascular malformations to avoid the increased intracerebral pressure and change of hemodynamic status during vaginal delivery$_{(12)}$, there is no established consensus on the mode of delivery. Especially cavernous malformations are low-blood flow lesions, and the potential hemorrhage associated with Valsalva maneuvers in vaginal deliveries is less probable(11), there is no strong scientific evidence supporting the selection of C-section.

Conclusion:

Symptoms such as syncope, dizziness, or headache are common in postpartum periods. In order to optimize postpartum care, primary care providers need skills to identify worrisome signs related to intracranial hemorrhage and arrange early diagnostic examinations, referral and treatment by a specialist. In patients diagnosed to have an intracranial vascular malformation in this period of time, it is vital to offer education about future bleeding risks, treatment indication and options. The selection of delivery mode should be based on general obstetric indications.

References

1. Fairhall JM, Stoodley MA. Intracranial haemorrhage in pregnancy. Obstet Med. 2009;2(4):142-8.
2. Maged AM, Mosaad M, AbdelHak AM, Kotb MM, Salem MM. The effect of hyoscine butylbromide on the duration and progress of labor in primigravidae: a randomized controlled trial. J Matern Fetal Neonatal Med. 2018;31(22):2959-64.
3. Klein AM, Loder E. Postpartum headache. Int J Obstet Anesth. 2010;19(4):422-30.
4. Sharma KJ, Kilpatrick SJ. Postpartum Hypertension: Etiology, Diagnosis, and Management. Obstet Gynecol Surv. 2017;72(4):248-52.

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5. Aronowski J, Zhao X. Molecular pathophysiology of cerebral hemorrhage: secondary brain injury. Stroke. 2011;42(6):1781-6.
6. Idiculla PS, Gurala D, Philipose J, Rajdev K, Patibandla P. Cerebral Cavernous Malformations, Developmental Venous Anomaly, and Its Coexistence: A Review. Eur Neurol. 2020;83(4):360-8.
7. Aoki R, Srivatanakul K. Developmental Venous Anomaly: Benign or Not Benign. Neurol Med Chir (Tokyo). 2016;56(9):534-43.
8. Cisneros O, Rehmani R, Garcia de de Jesus K. Cerebellar Cavernous Malformation (Cavernoma): A Case Report. Cureus. 2019;11(4):e4371.
9. Rabinstein AA, Flemming KD. Cavernous malformations with DVA: Hold those knives. Neurology. 2020;95(1):13-4.
10. Chen B, Herten A, Saban D, Rauscher S, Radbruch A, Schmidt B, et al. Hemorrhage from cerebral cavernous malformations: The role of associated developmental venous anomalies. Neurology. 2020;95(1):e89-e96.
11. Joseph NK, Kumar S, Brown RD, Jr., Lanzino G, Flemming KD. Influence of Pregnancy on Hemorrhage Risk in Women With Cerebral and Spinal Cavernous Malformations. Stroke. 2021;52(2):434-41.
12. Yamada S, Nakase H, Nakagawa I, Nishimura F, Motoyama Y, Park YS. Cavernous malformations in pregnancy. Neurol Med Chir (Tokyo). 2013;53(8):555-60.
13. Byrne JV. Cerebrovascular malformations. European Radiology. 2005;15(3):448-52.
14. Derdeyn CP, Zipfel GJ, Albuquerque FC, Cooke DL, Feldmann E, Sheehan JP, et al. Management of Brain Arteriovenous Malformations: A Scientific Statement for Healthcare Professionals From the American Heart Association/American Stroke Association. Stroke. 2017;48(8):e200-e24.
15. Ferrara AR. Brain arteriovenous malformations. Radiol Technol. 2011;82(6):543MR-56MR.
16. Miller TR, Gandhi D. Intracranial Dural Arteriovenous Fistulae: Clinical Presentation and Management Strategies. Stroke. 2015;46(7):2017-25.
17. Kashefiolasl S, Bruder M, Brawanski N, Herrmann E, Seifert V, Tritt S, et al. A benchmark approach to hemorrhage risk management of cavernous malformations. Neurology. 2018;90(10):e856-e63.