Pregnancy Combined with Epilepsy and Cerebral Cavernous Malformation

Ya-Lan Xu¹, Jun-Tao Liu¹, Yi-Jun Song¹, Xi-Ya Zhou¹, Qing-Wei Qi¹, Xu-Ming Bian¹, Zhi-Qin Xu², Lei Li²

¹Department of Obstetrics and Gynecology, Peking Union Medical College Hospital, Peking Union Medical College, Chinese Academy of Medical Sciences, Beijing 100730, China

²Department of Neurosurgery, Peking Union Medical College Hospital, Peking Union Medical College, Chinese Academy of Medical Sciences, Beijing 100730, China

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**INTRODUCTION**

Cerebral cavernous malformation (CCM), also known as cavernous angioma, is a type of vascular malformation in the central nervous system. To date, rare cases of CCM and seizure have been reported in pregnancies. Nowadays, the management of such condition in pregnancy is still a challenge due to lacking of experiences. We presented a case of pregnant woman with CCM combined with epilepsy and cerebral cavernous hemangioma.

**CASE REPORT**

A 31-year-old G1P1 woman presented to the Department of Obstetrics and Gynecology in Peking Union Medical College Hospital due to seizure at a gestational age of 36+4 weeks. She presented paroxysmal and abdominal seizure during sleep accompanied with white foam from the mouth and opisthotonos. No obvious seizure was observed in the limbs. The abdominal seizure was relieved 1–2 min later. The symptoms were presented again 3 h later, which were manifested as consciousness disorders, as well as strong tremor of four limbs. No eyeball turnover, tongue bite, urinary, and stool incontinence were observed. Ten minutes later, the consciousness was recovered and speech was normal. However, the patient showed dizziness and headache after seizure.

On physical examination, no aberrant changes were noticed. Monitoring on the fetal heart, movement, and uterine contraction was normal. Cranial magnetic resonance imaging (MRI) showed slightly long T1 and T2 signs (size: 2.0 cm × 2.1 cm × 1.5 cm) in the left frontal lobe, in which small punctiform low signal shadow was visible and low signal band was noted at the border. Fluid-attenuated inversion recovery showed high signal, and diffusion-weighted imaging showed isointensity. No obvious edema was found in adjacent brain parenchyma. The adjacent cortical sulci were not significantly widened [Figure 1]. The patient was finally diagnosed with intracranial space-occupying lesions and cavernous hemangioma. On this basis, the patient underwent uterine-incision delivery using combined spinal–epidural anesthesia on June 24, 2013, after signing the informed consent. The infant was healthy with an Apgar score of 10. After discharge, the patient showed no relapse of seizure. The infant was well developed during the follow-up. Intermittent seizure was present 2 years after the delivery. Therefore, surgical treatment was recommended by the neurosurgical physicians, but the patient did not receive the surgery due to thyroid cancer 1 year after delivery.

**DISCUSSION**

CCM was one of the important factors causing intracranial hemorrhage during gestational period. The incidence...
of intracranial hemorrhage was increased together with increase of cerebral angioma size in the pregnancy, which may be associated with the following aspects: the elevation of estrogen and progestogen resulted in high expression of growth factors (e.g., vascular endothelial growth factor and placenta growth factor), as well as change of blood pressure and proliferation of endothelial cells.[1]

Currently, there is no consensus on the selection of delivery mode for pregnant women with CCM. No Cochrane evidence revealed the fact that cesarean delivery was related with reduced incidence of intracranial hemorrhage. Meanwhile, no evidence indicated that the mode of delivery was associated with the risk of intracranial hemorrhage in CCM.[2] However, the incidence of hemorrhage in pregnancy with CCM was higher after receiving cesarean delivery or painless delivery at a gestational age of 32+ weeks (fetal weight >2 kg).[3] Therefore, interdepartmental cooperation is needed to select the delivery mode and decide whether neurosurgical procedures are needed to ensure the safety of pregnant women and the fetus. In this case, the patient showed seizure twice and was highly suspected with cavernous hemangioma after MRI and consultation. Considering the fetal age and the potential side effects of seizure on the child, cesarean delivery was given, and no relapse was noticed after delivery.

Up to now, there is no consensus on the treatment of CCM combined with seizure in pregnancy. Administration of certain drugs such as nimodipine has been proposed to attenuate vasospasm, but it may induce fetal deformity. Up to now, there are still disputes for the treatment efficiency of surgery. Lynch et al.[4] proposed that surgery was essential for the pregnancy with seizure or CCM. However, Kalani and Zabramski[5] reported that conservative therapy may be an option for the patients with mild or symptom-free CCM. Besides, the neurosurgery should be carried out according to the type of bleeding and severity of the conditions. In this study, the patient was recommended to receive surgery according to patient’s conditions. However, the surgery was refused due to the presence of thyroid cancer 1 year after delivery.

In conclusion, there is no increased risk of cranial hemorrhage associated with mode of delivery in the pregnancy with CCM. In the presence of hemorrhage, intervention should be given according to the type of hemorrhage.

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Conflicts of interest
There are no conflicts of interest.

References
1. Dias MS, Sekhar LN. Intracranial hemorrhage from aneurysms and arteriovenous malformations during pregnancy and the puerperium. Neurosurgery 1990;27:855-65.
2. Witiw CD, Abou-Handen A, Kulkarni AV, Silvaggio JA, Schneider C, Wallace MC. Cerebral cavernous malformations and pregnancy: Hemorrhage risk and influence on obstetrical management. Neurosurgery 2012;71:626-30. doi: 10.1227/NEU.0b013e31825fd0dc.
3. Burkhardt JK, Bozinov O, Nürnberg J, Shin B, Woernle CM, Ulrich NH, et al. Neurosurgical considerations on highly eloquent brainstem cavernomas during pregnancy. Clin Neurol Neurosurg 2012;114:1172-6. doi: 10.1016/j.clineuro.2012.02.040.
4. Lynch JC, Andrade R, Pereira C. Intracranial hemorrhage during pregnancy and puerperium: Experience with fifteen cases. Arq Neuropsiquiatr 2002;60:264-8. doi: 10.1590/S0004-282X2002000200015.
5. Kalani MY, Zabramski JM. Risk for symptomatic hemorrhage of cerebral cavernous malformations during pregnancy. J Neurosurg 2013;118:50-5. doi: 10.3171/2012.8.JNS12241.