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Corresponding Author

Rumi Bhattacharjee,
Department of Obstetrics and Gynaecology, Pramukhswami Medical College, Bhaikaka University, Karamsad, Anand, Gujarat, India

Author Designation

¹Professor
^{2,4}Assistant Professor
³3rd year resident
^{5,6}Third Year
⁷Senior Resident

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Pregnancy Complicated by a Ruptured Cerebral Arteriovenous Malformation: Unique Presentation and Tailored Management Approach: A Case Report

¹Rumi Bhattacharjee, ²Sangita Pandey, ³Sukhram Rathva, ⁴Shibashish Bhattacharjee, ⁵Jahnvi Patel, ⁶Jaimini Kapadia and ⁷Rashmita Pal

^{1,2,3,6,7}*Department of Obstetrics and Gynaecology, Pramukhswami Medical College, Bhaikaka University, Karamsad, Anand, Gujarat, India*

⁴*Department of Neurosurgery, Pramukhswami Medical College, Bhaikaka University, Karamsad, Anand, Gujarat, India*

ABSTRACT

Pregnancy represents a distinctive physiological state, where any cerebrovascular event can lead to various clinical consequences. Arteriovenous malformations (AVMs), abnormal connections between blood vessels, are primarily congenital and can vary in size and location. These malformations may remain asymptomatic for extended periods. However, rupture often occurs as a result of hemodynamic changes and can manifest as a stroke during pregnancy. Neurocritical complications associated with pregnancy have the potential to rapidly worsen without warning, underscoring the importance of vigilant monitoring and timely intervention for these patients. We present the case of a 26-year-old gravida 4 para 2 who developed cerebrovascular stroke at 27 weeks gestation consequent to a ruptured AVM and intracranial bleed but was managed conservatively till 35 weeks, after which she underwent an uneventful elective cesarean section, emphasizing both the uniqueness of the presentation and the individualized care provided.

INTRODUCTION

Cerebral arteriovenous malformations (CAVMs) are rare, inherited vascular anomalies with a prevalence ranging from 0.001%-0.5%. These malformations may present as isolated lesions in individuals without a family history or as part of familial forms linked to heterozygous loss-of-function mutations^[1]. CAVM are characterized by a central tangle of abnormally formed blood vessels, where feeding arteries merge or fuse, and the typical capillary network is absent^[2]. This results in a direct flow of oxygenated blood into the venous system, bypassing the capillary exchange. Consequently, the veins are subjected to high-pressure conditions, disrupting the delicate balance of cerebral circulation^[3]. Structurally, CAVMs resemble angiomas, encased by a wall of endothelial tissue devoid of connective tissue or muscle. The prevalence of these malformations is higher in women, particularly between the ages of 30 and 39 and they account for 10-15% of all vascular malformations within the central nervous system (CNS)^[4,5]. Clinically, CAVMs can present with a range of symptoms, including headaches, seizures, altered consciousness, or focal neurological deficits. A particularly concerning complication is intracranial hemorrhage (ICH), which during pregnancy is responsible for 8-38% of all cases of ICH^[6]. The diagnosis and management of CAVMs heavily rely on digital subtraction angiography (DSA), which remains the gold standard imaging technique for accurate diagnosis and treatment planning^[2]. Given the potential for significant neurological impairment and hemorrhagic risk, particularly in women of childbearing age, early detection and careful management are critical in improving outcomes for individuals affected by AVMs.

Case Description: A 26-year-old woman, G4P2A1L2, with a history of cerebrovascular stroke at 27.6 weeks gestational age (GA), was managed conservatively in the ICU at a private facility following the stroke. During the episode, she had a seizure and has since been on antiepileptic medications. Initial MRI findings were suggestive of an arteriovenous malformation (AVM), Spetzler-Martin grade 111, complicated by acute intraparenchymal hemorrhage with intra ventricular extension of bleed involving the frontal and occipital horns of the bilateral lateral ventricles, third ventricle, and fourth ventricle. The lesion size was 2.8 cm with a volume of 9.8 cm (Fig 1). Her previous two vaginal deliveries at term were uncomplicated. She presented at 35.4 weeks GA in the emergency center of our facility with preterm labor pains. On admission, she was vitally stable with no residual limb weakness on neurological examination. A follow-up MRI showed a

reduction in the size of the intraparenchymal and intraventricular hemorrhages and a decrease in mass effect and midline shift (Fig 2). A neurosurgical consultation was obtained and the patient and her family were provided with comprehensive information about the available obstetric management options, including the potential risks and benefits. After a thorough discussion, a multidisciplinary team decided on an elective lower-segment cesarean section (LSCS). The patient underwent an elective LSCS with tubal ligation at 35.6 weeks GA under general anesthesia. She delivered a healthy 2.4 kg infant without any maternal or fetal complications. Her postoperative course was uneventful and she was discharged on antiepileptic medications. Upon discharge, the patient and her relatives were counseled on the need for definitive surgical management of the AVM. Three months postpartum, a follow-up found the patient in stable condition and a surgical procedure was advised to address the AVM and prevent further complications.

RESULTS AND DISCUSSIONS

Intracranial hemorrhage (ICH) and stroke are recognized as the most common clinical manifestations of cerebral arteriovenous malformations (CAVMs) during pregnancy. Aoyama *et al.* highlighted pregnancy as a significant risk factor for stroke, with approximately 60% of all strokes occurring during pregnancy and the puerperium being of the hemorrhagic type^[7]. A study by Vest *et al.*, which examined the etiology of ICH during pregnancy and the puerperium using the SMASH-U classification (structural vascular lesion, medication, amyloid angiopathy, systemic disease, hypertension, undetermined), found that 31.3% of ICH cases were linked to structural lesions, with CAVMs accounting for 18.4%^[8]. Notably, structural lesions were more prevalent during pregnancy, underscoring the heightened risk during this period. The turbulent, high-velocity and low-resistance blood flow within a CAVM bypasses the cerebral hemodynamic mechanisms of autoregulation^[9]. It is postulated that the physiological hemodynamic changes during pregnancy contribute to increased distension within the already tortuous vascular networks of the AVM and induce subtle damage at the microvascular level. These changes serve to exacerbate pre-existing pressure symptoms, intensifying the overall clinical presentation^[10]. It is believed that chronic headaches, epilepsy and progressive neurological deficits are not typical features of cerebral arteriovenous malformations (CAVMs)^[3]. A non-ruptured AVM generally elicits subtle neurological symptoms, which are influenced primarily by the location and size of the

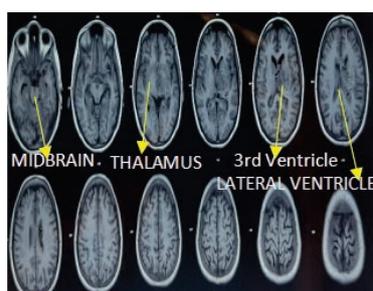


Fig 1: MRI Brain

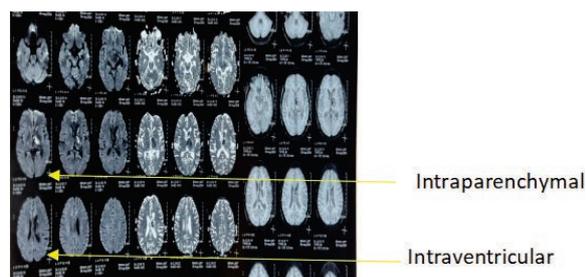


Fig 2: MRI Brain

lesion. Still, these symptoms tend to be minimal unless the AVM is large. In contrast, rupture of the AVM often leads to more pronounced symptoms, including those related to peri-hematoma edema and the “mass effect,” which results from increased pressure and swelling in the surrounding brain tissue^[10]. Regarding the timing of rupture, both De Maria and Pohjola *et al.* have suggested a greater likelihood of rupture occurring during the second or third trimester of pregnancy^[2,11]. In a cohort study by Pohjola *et al.*, 43% of ruptures were observed in the second and 24% in the third trimester^[11]. The second-trimester rupture in our case mirrors the findings of Magitta *et al.*, while Kow *et al.* reported a subarachnoid hemorrhage at 31 weeks and 3 days of gestational age^[10,12]. However, other researchers, including Etter MM and Sappenfield *et al.*, have presented varying results regarding the risk of CAVM rupture during pregnancy^[13,14]. Our patient's age and initial symptomatology align with the findings of Yan *et al.*, who reported a mean age of 26.6 years, and Pohjola *et al.*, who cited a mean age of 28.9 years at presentation. Additionally, Kow *et al.* described cases involving multiparous women with previous uncomplicated vaginal deliveries, a profile that closely mirrors the obstetric history of our patient. The diagnostic modalities for cerebral arteriovenous malformations (CAVMs) include non-contrast CT, where CAVMs typically appear as a hyperdense mass., plain MRI, which reveals tangled flow voids and CT or MRI angiography, which provides detailed information on the location, size and feeding vessels of the nidus and any coexisting aneurysms^[15]. The choice of diagnostic modality during pregnancy must be carefully

considered, considering the patient's clinical condition, the situation's urgency and the differential diagnosis^[12]. For diagnosing intracranial hemorrhage (ICH) in pregnancy, a CT scan is generally preferred due to its ability to provide rapid results with minimal fetal radiation exposure (<0.005 mGy)^[12]. MRI, conversely, is practical for identifying the underlying cause. According to Maria *et al.*, the fetal radiation exposure associated with digital subtraction angiography (DSA) during pregnancy is minimal and considered an insignificant effective dose. The International Commission on Radiological Protection (ICRP) recommends that the maximum radiation dose to the pregnant uterus should not exceed 100 mGy. With the use of appropriate uterine shielding, the calculated fetal radiation dose during diagnostic and therapeutic endovascular procedures can be as low as 0.02 mGy, ensuring minimal exposure to the fetus^[16]. Definitive management options for a cerebral arteriovenous malformation (CAVM) include conservative medical treatment, microsurgical excision, stereotactic radiosurgery and endovascular embolization, which aims to reduce the size of the nidus or eliminate associated aneurysms, venous varices and fistulas^[9]. The Spetzler-Martin grading system is commonly used to assess surgical risk for AVMs^[9]. Ideally, surgical intervention should be performed before or during the early stages of pregnancy or the puerperium^[2]. When managing a ruptured AVM, intervention is typically guided by neurological indications rather than obstetric considerations^[16]. Surgery remains the cornerstone of management for cerebral arteriovenous malformations (CAVMs). However, Derdeyn *et al.* referenced the findings of the ARUBA trial, which suggested that medical management may be superior to surgical intervention in the case of unruptured CAVMs^[17]. Kow *et al.* and Magitta *et al.* have also applied this approach in managing ruptured CAVMs and reported favorable outcomes, a course observed in our case. In the case presented by Kow *et al.*, a subarachnoid hemorrhage at 31 weeks and 3 days of gestational age was managed conservatively. The patient remained neurologically stable and an elective cesarean section was performed at 34 weeks and 5 days, followed by definitive surgery on the sixth postpartum day. Similarly, Magitta *et al.* chose conservative management for an early second-trimester rupture of bilateral parietal AVMs. The pregnancy progressed well, with complete resolution by 7 months gestational age and the patient delivered at term without complications, resulting in a healthy baby. In obstetric management, the timing of delivery must carefully balance the risks of prematurity against the potential for re-bleeding during pregnancy. Kow *et al.* report a

27-30% risk of re-bleeding during the same pregnancy, in contrast to a significantly lower risk of 2-6% annually in the nonpregnant population^[12]. Asano estimates the re-bleeding risk at approximately 27%, while Pohjola *et al.* highlight an increased risk of initial rupture during vaginal delivery. These considerations underscore the importance of individualized decision-making when managing CAVMs during pregnancy. Regarding the mode of delivery, Teik *et al.* advocate for an elective cesarean section (LSCS), citing its ability to provide better control over hemodynamic stability^[13]. In contrast, Kow *et al.* argue that a cesarean section avoids the physiological changes in intracranial pressure associated with uterine contractions. In their study of ruptured CAVMs during pregnancy, Yan *et al.* reported that 69% of patients who underwent emergent intervention for the AVM before delivery had cesarean sections^[6]. However, Etter *et al.*, in their systematic review, noted that the risk of adverse cerebrovascular events is higher following a cesarean section compared to vaginal delivery, highlighting the need for careful consideration of delivery methods in these cases^[13]. Pohjola *et al.* suggest that the final decision regarding the mode of delivery in unruptured CAVM cases may largely depend on the preference of the patient or couple. However, vaginal delivery in the case of a ruptured CAVM could present significant challenges. Despite these risks, Etter *et al.* have reported favorable maternal-fetal outcomes in ruptured CAVMs, both in their case series and literature review, underscoring the possibility of a positive outcome with careful management^[11-14].

CONCLUSION

A ruptured CAVM during pregnancy requires prompt diagnosis and a collaborative approach involving obstetricians, neurologists, neurosurgeons and interventional neuroradiologists. Our present case report emphasizes the importance of this multidisciplinary teamwork, where coordinated efforts and tailored obstetric care and counseling facilitated the development of an individualized management plan, leading to an optimal maternal and fetal outcome.

REFERENCES

- Al-Shahi, R., J.J. Bhattacharya, D.G. Currie, V. Papanastassiou and V. Ritchie *et al.*, 2003. Prospective, Population-Based Detection of Intracranial Vascular Malformations in Adults. *Stroke*, 34: 1163-1169.
- Maria, L.D., S. Seriola and M.M. Fontanella, 2023. Brain Arteriovenous Malformations and Pregnancy: A Systematic Review of the Literature. *World Neurosurg.*, 177: 100-108.
- Florian, I., L. Beni, V. Moisiu, T. Timis, I. Florian, A. Bala?a and I. Berindan-Neagoe, 2021. 'De Novo' Brain AVMs—Hypotheses for Development and a Systematic Review of Reported Cases. *Medicina*, Vol. 57 .10.3390/medicina57030201.
- Merlino, L., F.D. Prete, L. Titi and M.G. Piccioni, 2020. Cerebral cavernous malformation: Management and outcome during pregnancy and puerperium. A systematic review of literature. *J. Gynecol. Obstet. Hum. Reprod.*, Vol. 50 .10.1016/j.jogoh.2020.101927.
- Taslimi, S., A. Modabbernia, S. Amin-Hanjani, F.G. Barker and R.L. Macdonald, 2016. Natural history of cavernous malformation. *Neurology*, 86: 1984-1991.
- Yan, K.L., N.U. Ko, S.W. Hetts, S. Weinsheimer, A.A. Abal, M.T. Lawton and H. Kim, 2021. Maternal and Fetal Outcomes in Women with Brain Arteriovenous Malformation Rupture during Pregnancy. *Cerebrovascular Dis.*, 50: 296-302.
- Aoyama, K. and J.G. Ray, 2020. Pregnancy and Risk of Intracerebral Hemorrhage. *JAMA Network Open*, Vol. 3 .10.1001/jamanet work open.2020.2844.
- Vest, T., K. Rantanen, L. Verho, K. Aarnio and A. Korhonen *et al.*, 2024. Etiology of intracerebral hemorrhage during pregnancy or puerperium: A nationwide study. *Eur. J. Neurol.*, Vol. 31 .10.1111/ene.16012.
- Peschillo, S., A. Caporlingua, C. Colonnese and G. Guidetti, 2014. Brain AVMs: An Endovascular, Surgical and Radiosurgical Update. *Sci. World J.*, Vol. 2014 .10.1155/2014/834931.
- Magitta, N.F., E.M. Sindato, J.R. Meda, H.N. Toroha and A.J. Meremo, 2023. Ruptured bilateral brain arteriovenous malformations in a young woman with early pregnancy: A case report. *J. Med. Case Rep.*, Vol. 17 .10.1186/s 13256-023-03946-6.
- Pohjola, A., T. Vest, L. Verho, K. Aarnio and K. Rantanen *et al.*, 2024. Intracranial Arteriovenous Malformations During Pregnancy and Puerperium-A Retrospective Nationwide Population-Based Cohort Study. *Neurosurgery*, Vol. 96 .10.1227/neu.0000000000003067.
- Kow, C.S., L. Yang, W.C. Tan, R.W.L. Leong, Y.P. Ng, S. Arunachalam and D. Kanagalingam, 2022. An Unusual Case of Cerebral Arteriovenous Malformation in Pregnancy. *J. Med. Cases*, 13: 104-108.
- Etter, M.M., A. Nguyen, A. Brehm, C. Aberle and I. Tsogkas *et al.*, 2023. Endovascular Treatment and Peri-interventional Management of Ruptured Cerebrovascular Lesions During Pregnancy. *Clin. Neuroradiology*, 33: 833-842.

14. Sappenfield, E.C., R.T. Jha, S. Agazzi and S. Ros, 2019. Cerebral arteriovenous malformation rupture in pregnancy. *BMJ Case Rep.*, Vol. 12 .10.1136/bcr-2018-225811.
15. Teik C.K. and N.L. Basri *et al.*, 2019. Management Options and Outcome of Cerebral Arteriovenous Malformation in Pregnancy: Case Series. *Arch Iran Med.*, 22: 340-343.
16. Asano, S., N. Hayashi, S. Edakubo, M. Hosokawa and J. Suwa *et al.*, 2016. Successful perinatal management of a ruptured brain arteriovenous malformation in a pregnant patient by endovascular embolization followed by elective cesarean section: A single-case experience. *JA Clin. Rep.*, Vol. 2 .10.1186/s40981-016-0045-6.
17. Derdeyn, C.P., G.J. Zipfel, F.C. Albuquerque, D.L. Cooke, E. Feldmann, J.P. Sheehan and J.C. Torner, 2017. Management of Brain Arteriovenous Malformations: A Scientific Statement for Healthcare Professionals From the American Heart Association/American Stroke Association. *Stroke*, 48: 200-224.