Case Report

Successful Cesarean Section Deliveries in a Patient with a History of Developmental Venous Anomaly-Induced Hemorrhage

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While hemorrhage can occur because of developmental venous anomalies (DVAs), there is no established opinion concerning their association with pregnancy and childbirth. In the present report, we discuss the case of a now 39-year-old woman with DVA in whom pregnancy and childbirth were successful. When she was 28, she experienced disturbance of consciousness and paralysis on the left side of the body, and brain computed tomography revealed cerebral hemorrhage coupled with subarachnoid hemorrhage. Cerebral angiography revealed a DVA with an arteriovenous shunt, with superficial drainage surrounding the hematoma. No associated cavernous hemangiomas were observed, and the patient was diagnosed with DVA-induced hemorrhage and treated via conservative therapy. Later, at the ages of 32 and 35, she gave birth via Caesarean section under general anesthesia. At the age of 37, she experienced sudden headache and nausea, following which she was again diagnosed with DVA-induced hemorrhage. Fortunately, she experienced no exacerbation of symptoms such as paralysis. However, she currently has mild, residual paralysis on the left side of the body, and she regularly walks to the hospital using a cane for follow-up examinations.

Key Words: Developmental venous anomaly—intracranial hemorrhage—pregnancy—childbirth

Introduction

Developmental venous anomaly (DVA), formerly known as venous angioma, is the most prevalent type of cerebral vascular malformation. DVA plays a role in normal cerebral venous return and is essentially benign and asymptomatic, although it has also been associated with hemorrhage derived from cavernous malformations. Moreover, in rare cases, hemorrhage may occur due to DVA itself, having been reported in a pediatric case[1] and during pregnancy.[2-5] A recent report revealed that DVA can occur in combination with arteriovenous (AV) shunts, the presence of which is believed to affect the rate of hemorrhage.[6]

It remains controversial whether pregnancy and childbirth should be advised in patients with DVA. While previous research reported that the hemorrhage rate is elevated during pregnancy,[2,3] more recent studies have argued that DVA does not constitute an adequate ground for avoiding pregnancy in patients without substantial hemorrhage.[7] However, the risk of pregnancy and

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childbirth in patients with a history of hemorrhage or AV shunts remains unclear. We consider the present case extremely valuable for assessing the safety of pregnancy and childbirth in patients with similar symptoms.

Case Report

We discuss the case of a now 39-year-old woman who underwent brain computed tomography (CT) at the age of 28 due to impaired consciousness and paralysis on the left side of the body. Computed tomography revealed that she had experienced a brain hemorrhage extending from the right temporal lobe to the putamen, which was complicated by a subarachnoid hemorrhage surrounding a hematoma. Cerebral angiography revealed a DVA with superficial drainage surrounding the hematoma. Medullary veins extending from the arterial phase were coupled with AV shunts. She was diagnosed with brain hemorrhage associated with DVA, following which she underwent conservative therapy. During hospitalization, she was found to be in early pregnancy, and the pregnancy was terminated. Despite mild, residual paralysis on the left side of the body, her consciousness improved, and she was discharged home. Cerebral angiography was performed 1 month (Fig 1) and 1 year after onset, revealing no changes in her hemodynamic status. During follow-up, MRI confirmed that the hematomas and associated cavernous hemangiomas had resolved, indicating that the hemorrhage had been derived from the DVA itself. Several years later, at the ages of 32 and 35, she gave birth via Caesarian section under general anesthesia. At the age of 37, she experienced sudden headache and nausea, following which she was again diagnosed with DVA-induced hemorrhage. Cerebral angiography revealed no noticeable changes from the first DVA onset. Fortunately, she experienced no exacerbation of symptoms such as paralysis due to the second hemorrhage incident, and she currently visits the outpatient clinic on a regular basis for follow-up examinations.

Discussion

The present case is characterized by 3 features: (1) The patient was pregnant during the first hemorrhage incident, (2) she experienced 2 successful deliveries despite a history of hemorrhage, and (3) hemorrhage recurred independently of her pregnancy status.

In the past, pregnancy and childbirth were believed to increase the risk of DVA-induced hemorrhage.[2,3] However, recent reports have mentioned that hemorrhage from DVAs during pregnancy and childbirth were caused by outflow obstruction due to clot formation in DVA during lochia[4] or increased coagulability during the puerperal period.[5] In the present case, MRI and cerebral angiography were performed immediately after the 2 bleeding episodes; however, we were unable to detect evidence of thrombosis within DVA. Moreover, her prothrombin time and activated partial thromboplastin time showed no abnormalities. Unfortunately, we did not perform other tests for the detection of abnormalities in the coagulation system, such as tests for d-dimer, antithrombin III, and protein C, among others. In 1 previously reported case, the patient experienced perimesencephalic subarachnoid hemorrhage following a Valsalva maneuver, an act similar to the urge to push during delivery.[8] Angiography revealed DVA enlargement in another patient,[9] indicating that pregnancy and childbirth may be indirect risk factors for hemorrhage. Such reports have gradually uncovered the mechanisms underlying DVA-associated hemorrhage during pregnancy or childbirth, and it is unlikely that pregnancy and childbirth directly increase the risk of hemorrhage from DVA. In the present case, AV shunts may have been responsible for the first hemorrhage incident. Im et al reported that 8 of 15 patients with DVA accompanied by AV shunts developed hemorrhage.[6] Moreover, hemorrhage recurred in 3 patients in whom AV shunts appeared in the early phase of angiography; this finding suggested that the early appearance of AV shunts increases the risk of hemorrhage in patients with DVA. While no previous studies have reported an association between pregnancy and AV shunts, our findings suggest that hemorrhage risk is increased in pregnant patients with AV shunts.

Rubin et al discussed the successful deliveries of 2 women with DVA without hemorrhage. Based on a review of the literature, the authors concluded that pregnancy and childbirth are safe in patients with DVA without hemorrhage.[7] While the risk of pregnancy and childbirth in patients with DVA-induced hemorrhage remains debatable, our patient experienced 2 successful deliveries despite a history of hemorrhage. Similarly, Malik et al reported the case of a patient who experienced hemorrhage during the second trimester of pregnancy, who later became pregnant again and delivered a child.[3] Taken together, these findings suggest that a history of hemorrhage may not warrant termination of the pregnancy.

Our patient experienced hemorrhage recurrence 9 years after the first hemorrhage incident. Given that DVA-associated hemorrhage is an extremely rare phenomenon, we speculate that AV shunts increased the risk of hemorrhage in our patient.

Patients with DVA without a history of hemorrhage generally experience safe pregnancies and childbirth.[7] However, the risks associated with pregnancy and childbirth in patients with a history of hemorrhage or AV shunts remain unclear. The findings from the present case suggest that the presence of an AV shunt or a history of hemorrhage are risk factors for hemorrhage during

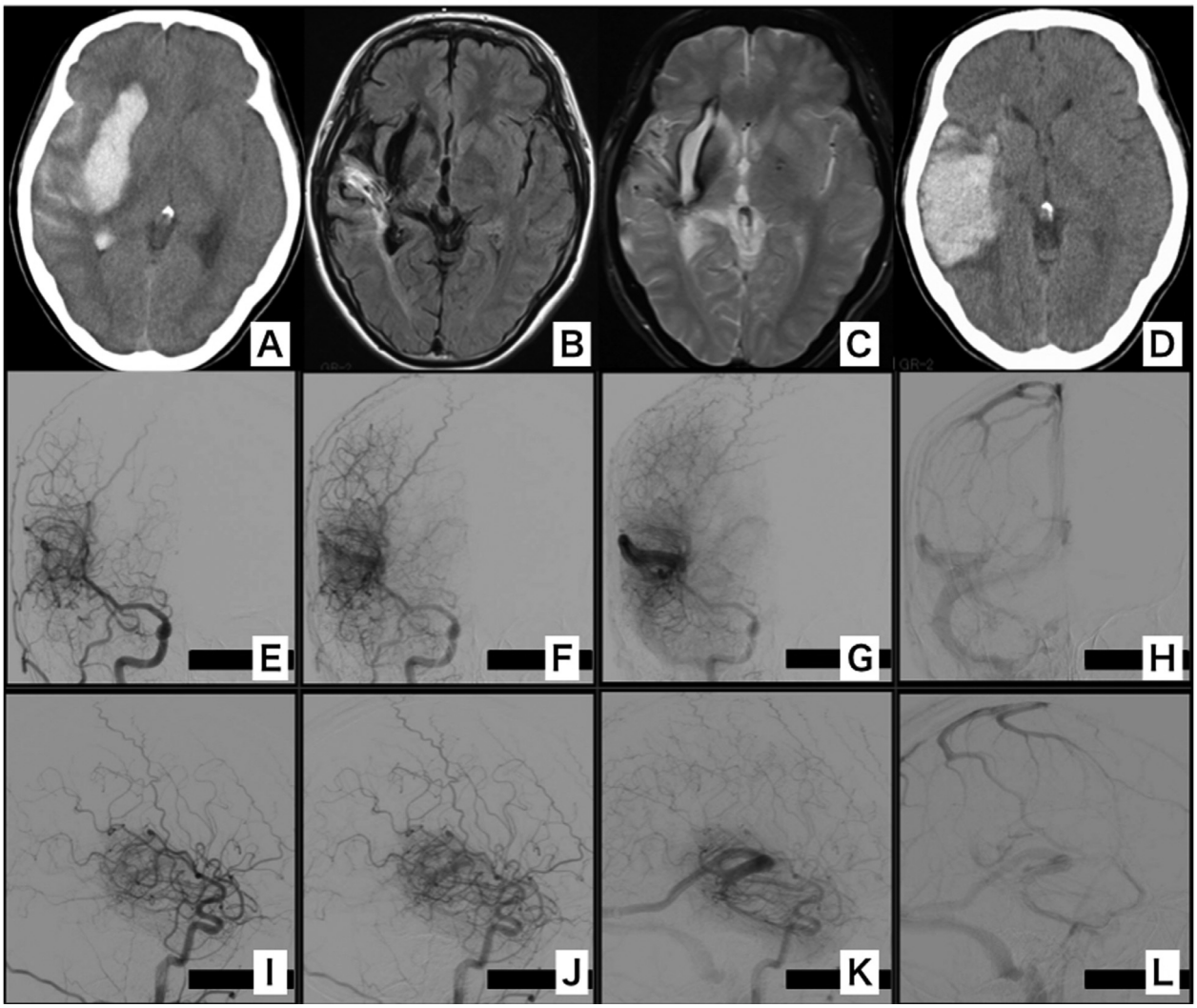

Figure 1. *Brain computed tomography (CT) (A) at 28 years of age (first hemorrhage incident): brain and subarachnoid hemorrhages extended from the right putamen to the temporal subcortical region. Cerebral angiography 1 month after onset: images of the right common carotid artery (frontal view: E-H, lateral view: I-L) revealed the capillary brush via the cortical branch of the middle cerebral artery in the early arterial phase (E, I) and mid-arterial phase (F, J). Outflow to the vein of Labbe was observed via the medullary vein, which extended to the late arterial phase (G, K). Other normal drainage pathways including the vein of Trolard and the superficial Sylvian vein were noted in the venous phase (H, L). Vertebral arteriography and left common carotid arteriography revealed no marked anomalies. T2*-weighted fluid-attenuated inversion recovery magnetic resonance images (FLAIR-MRI) (B) during follow-up. (C) Images revealed that the hematoma had resolved, although hemosiderosis was observed at the original bleeding site. No cavernous malformations were noted. Brain CT (D) at 37 years of age (second hemorrhage incident): while a gigantic hematoma was observed in the right temporal subcortical region, the hemorrhage filled the space within the brain parenchyma that had already atrophied, and the mass effect on normal brain tissue was small.*

pregnancy or childbirth in patients with DVA. Such patients should undergo cesarean section in order to prevent lochia, which may induce thrombus formation, and avoid the pushing required during vaginal delivery.

Disclosure

The authors report no conflicts of interest concerning the materials or methods used in this study or the findings specified in this paper.

References

1. Entezami P, Boulos A, Yamamoto J, et al. Paediatric presentation of intracranial haemorrhage due to thrombosis of a developmental venous anomaly. BMJ Case Rep 2019;12:e227362.
2. Garner TB, Del Curling Jr. O, Kelly Jr. DL, et al. The natural history of intracranial venous angiomas. J Neurosurg 1991;75:715-722.
3. Malik GM, Morgan JK, Boulos RS, et al. Venous angiomas: an underestimated cause of intracranial hemorrhage. Surg Neurol 1988;30:350-358.

4. Seki M, Shibata M, Itoh Y, et al. Intracerebral hemorrhage due to venous thrombosis of developmental venous anomaly during pregnancy. J Stroke Cerebrovasc Dis 2015;24:e185-e187.
5. Yamamoto M, Inagawa T, Kamiya K, et al. Intracerebral hemorrhage due to venous thrombosis in venous angioma-case report. Neurol Med Chir (Tokyo) 1989;29:1044-1046.
6. Im SH, Han MH, Kwon BJ, et al. Venous-predominant parenchymal arteriovenous malformation: a rare subtype with a venous drainage pattern mimicking developmental venous anomaly. J Neurosurg 2008;108:1142-1147.
7. Rubin SM, Jackson GM, Cohen AW. Management of the pregnant patient with a cerebral venous angioma: a report of two cases. Obstet Gynecol 1991;78:929-931.
8. Ruíz DS, Delavelle J, Yilmaz H, et al. Parenchymal abnormalities associated with developmental venous anomalies. Neuroradiol 2007;49:987-995.
9. Saeed O, Khan AA, Herial NA, et al. Exercise induced transient neurological deficit in a patient with cerebellar developmental venous anomaly. J Vasc Interv Neurol 2015;8:17-20.