

Improvement of intrapartum reversible cerebral vasoconstriction syndrome over 12 weeks after onset: A Case Report

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Abstract

Background: We report a case of intrapartum reversible cerebral vasoconstriction syndrome and the improvement course of vasoconstriction over 12 weeks.

Case presentation: The patient was a 37-year-old woman without gestational hypertension. She entered labor at 40 weeks of gestation and presented 19 hours later with severe headache and tonic-clonic seizures. After the delivery of the fetus by emergency cesarean section, a computed tomography scan of the head showed cortical subarachnoid hemorrhage. Magnetic resonance imaging on the following day showed cerebral infarction in the right occipital lobe and thalamus. The next day, magnetic resonance angiography (MRA) showed mild vasoconstriction in the right posterior cerebral artery, which progressed from the peripheral to the central portion on day 5. MRA at week 12 and week 23 showed residual vasoconstriction in the right posterior cerebral artery, which was improved at week 39.

Conclusions: This is the first report demonstrating improvement of vasoconstriction with a long-term follow-up of more than 12 weeks by MRA.

Keywords: Reversible cerebral vasoconstriction syndrome; Perinatal cerebrovascular disease; delayed improvement; MRA in long term

Introduction

Reversible cerebral vasoconstriction syndrome (RCVS) is characterized by reversible intracranial blood vessel vasoconstriction and various intracranial lesions, such as convexity subarachnoid hemorrhage and ischemic lesions after severe headaches. [1] The vasoconstriction usually disappears within 12 weeks with a favorable prognosis, and no report

has demonstrated noticeable improvement of vasoconstriction with follow-up imaging for more than 12 weeks. [2] We report a case of RCVS that suddenly occurred during delivery and persisted for more than 12 weeks but improved after 39 weeks.

Case Report

The patient was a 37-year-old nulliparous woman without an episode of migraine and abnormal inflammatory disease. She was admitted to another hospital 40 weeks after the onset of labor. Nineteen hours later, she developed a severe bilateral temporal headache with a 2-minute tonic-clonic seizure. To treat eclampsia, magnesium sulfate was administered intravenously at a dose of 1 g per hour, and her consciousness improved once. However, after arriving at our emergency room, she had a tonic-clonic seizure again to be treated with diazepam 10 mg. Because the fetus showed prolonged bradycardia in electronic fetal heart rate monitoring, we conducted a cesarean section without a head imaging examination. After the delivery, she underwent a head computed tomography scan, and a cortical subarachnoid hemorrhage localized in the right frontal sulcus was confirmed (Figure 1A). On the second day

after admission, she complained of abnormal sensations on the left side and left hemianopsia.

Urgent contrast magnetic resonance imaging demonstrated new infarcts in the right occipital lobe and thalamus and angiogenic edema in the bilateral occipital lobes (Figure 1B, C). Magnetic resonance angiography (MRA) showed peripheral stenosis in the right posterior cerebral artery (PCA) (Figure 1D). MRA on the fifth day showed progression of the stenosis on the proximal side of the right P1 (Figure 1E). She was discharged from our hospital on the 19th day of taking the calcium channel blocker medication. The peripheral right PCA was not visualized on MRA at 23 weeks (Figure 1F) but became pictured at 39 weeks (Figure 1G).

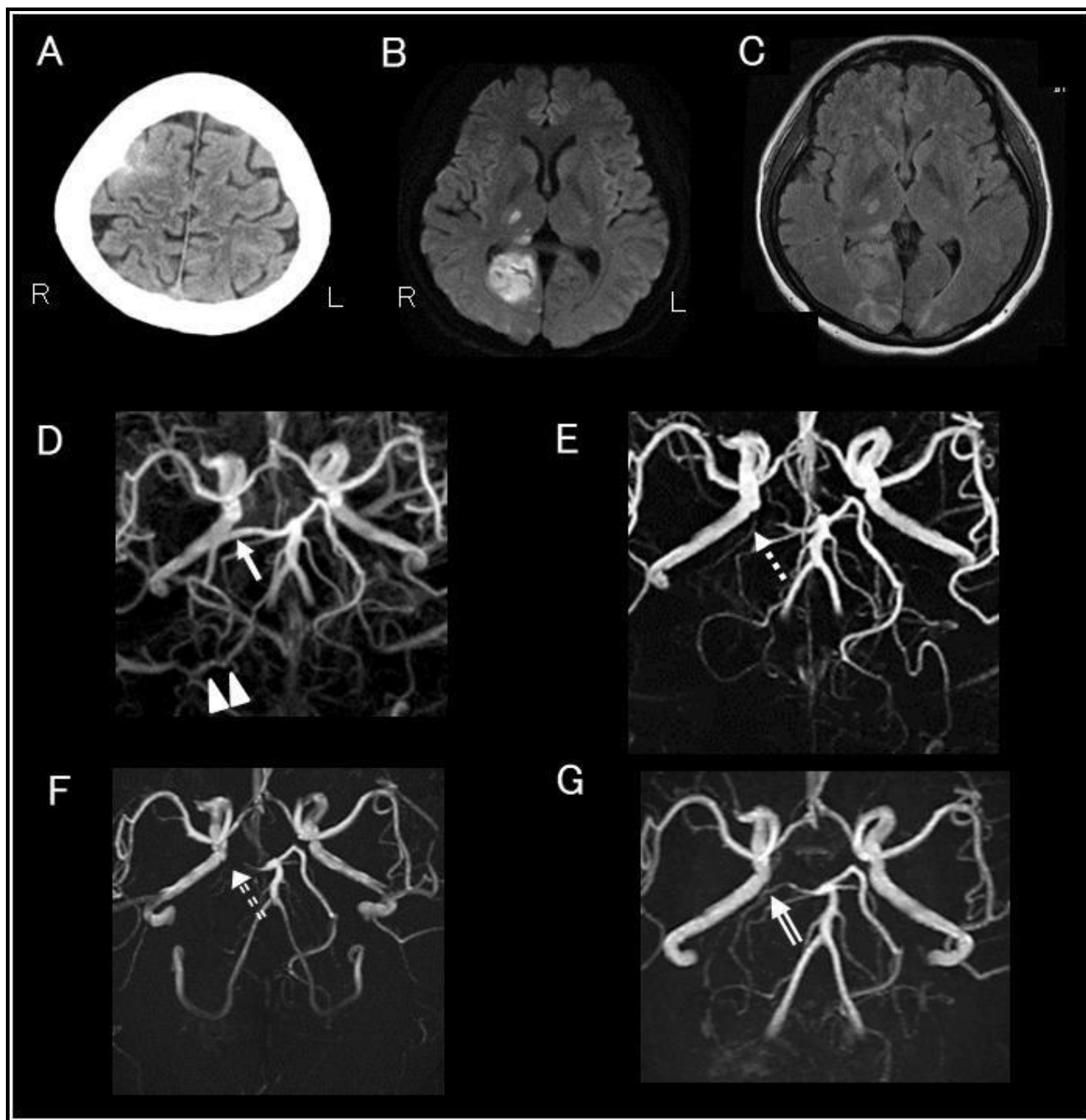


Figure 1: Head computed tomography on the first day (A) shows subarachnoid hemorrhage confined to the sulcus of the right frontal lobe. Head magnetic resonance imaging diffusion-weighted image (B) and Fluid Attenuated Inversion Recovery image (C) on the second day shows new infarction of the right occipital lobe and thalamus and angiogenic edema in the bilateral occipital lobes.

Head magnetic resonance angiography shows

(D) Second day: Two peripheral stenoses in the right posterior cerebral artery (PCA) are seen (Arrowhead: bead-like stenosis). No remarkable stenosis was depicted proximal PCA (Arrow).

(E) fifth day: Progressive vasoconstriction of the right PCA (Dotted arrow).

(F) Week 23: No recanalization of the right PCA (Dotted double arrow).

(G) Week 39: Vasoconstriction of the right PCA is improved (Double arrow).

Discussion

This is the first report showing intrapartum RCVS improved after 12 weeks. According to standard diagnostic criteria, vasospasm resolves within 12 weeks, but there are few descriptions of vasospasm lasting more than 12 weeks. [1,2] Our patient had multifocal areas of reversible constriction involving the cerebral arteries followed by acute onset of severe headache (“thunderclap headache”) that is considered characteristic of RCVS. [1] Since aneurysmal rupture, arterial dissection, and angiitis, listed in the differential diagnosis of RCVS, were negative, this case is considered an atypical RCVS with the delayed improvement of vasoconstriction. The cause of prolonged vasoconstriction is unknown, but it may be due to a combination of cortical subarachnoid

hemorrhage and pregnancy. One of the causes of protracted vasoconstriction has been reported to be the thickening of the vascular wall due to vascular inflammation due to subarachnoid hemorrhage. [3,1] Additionally, a study of severe RCVS in pregnant women reported that increased sensitivity to sympathomimetics due to hormonal fluctuations of pregnancy was associated with exacerbations.

[4] The prolonged vasoconstriction, in this case, might be caused by hormonal changes related to pregnancy combined with vascular damage and wall thickening due to subarachnoid hemorrhage. We need more points in the future to understand this condition.

The characteristics of vasoconstriction in RCVS were reported to begin in the

periphery and move to the center over time, followed by complete resolution of the vasoconstriction. [5] It has been reported that vasospasm may not be detected by cerebral angiography or MRA, even if parenchymal lesions appear in RCVS. [2] One reason is that vasospasm in early RCVS is peripheral and not detectable on imaging.

[5] In perinatal RCVS, there are also reports of poor prognosis that vasospasm was not observed in the first MRA and then appeared later than subarachnoid hemorrhage and cerebral infarction. [4] Therefore, considering the risk of performing MRI or cerebral angiography for rapid

Conclusions

RCVS should be considered in cases of severe headache followed by convex subarachnoid hemorrhage and cerebral edema, even in situations where angiography or MRI is not available in labor. RCVS is a reversible disease, but as this case shows, vasoconstriction can last for more than 12 weeks.

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Conflict of Interest: None

Abbreviations:

RCVS: Reversible cerebral vasoconstriction syndrome

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changes in the maternal body, as in this case, RCVS should be considered in patients with convex subarachnoid hemorrhage and cerebral edema.

Calcium channel blockers, magnesium sulfate, and steroids have been reported as treatments for RCVS, and the usefulness of magnesium sulfate has been reported in perinatal RCVS, which is often complicated by eclampsia, as in this case. [4] There is concern that calcium channel blockers may reduce placental blood flow due to maternal hypotension, and caution should be exercised when administering them before delivery.

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Availability of data and materials: The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request

Ethics approval and consent to participate: Consent was obtained from the patient. This study adhered to the tenets of the Declaration of Helsinki.

Consent for publication: Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Data availability: All data generated or analyzed during this study are included in this article. Further enquiries can be directed to the corresponding author.

References

1. Calabrese LH, Dodick DW, Schwedt TJ, Singhal AB (2007) Narrative review: reversible cerebral vasoconstriction syndromes. *Ann Intern Med.* 146(1): 34-44.
2. Ducros A (2012) Reversible cerebral vasoconstriction syndrome. *The Lancet Neurology.* 11(10): 906-917.
3. Borel CO, McKee A, Parra A, Haglund MM, Solan A, et al. (2003) Possible role for vascular cell proliferation in cerebral vasospasm after subarachnoid hemorrhage. *Stroke.* 34(2): 427-33.
4. Fugate JE, Wijdicks EF, Parisi JE, Kallmes DF, Cloft HJ, et al. (2012) Fulminant postpartum cerebral vasoconstriction syndrome. *Arch Neurol.* 69(1): 111-7.
5. Shimoda M, Oda S, Hirayama A, Imai M, Komatsu F, et al. (2016) Centripetal Propagation of Vasoconstriction at the Time of Headache Resolution in Patients with Reversible Cerebral Vasoconstriction Syndrome. *AJNR Am J Neuroradiol.* 37(9): 1594-8.