

Convexity Subarachnoid Haemorrhage During Childbirth Related to Reversible Cerebral Vasoconstriction Syndrome – A Case Report

Sulkalno subarahnoidno krvarenje tijekom poroda vezano uz sindrom reverzibilne cerebralne vazokonstrikcije – prikaz slučaja

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Abstract. Aim: Pregnancy-related subarachnoid haemorrhage (pSAH), while uncommon, carries a higher risk of morbidity and mortality. The aim of the case presentation is to emphasize the importance of promptly identifying neurological complications during pregnancy and the postpartum period. **Case report:** A 39 weeks pregnant secundipara gave birth to a healthy baby. Just before childbirth she started signalling a right sided thunderclap headache followed by a blurred vision. Brain non-contrast computed tomography (NCCT) showed a convexity subarachnoid haemorrhage (cSAH). CT angiography (CTA) and MR angiography (MRA) of the brain revealed a multifocal vasospasm involving few of the arteries of Circle of Willis and a case of reversible cerebral vasoconstriction syndrome (RCVS) was suspected. The patient was treated with nimodipine, and a follow up three months after the event showed the complete resolution of the cSAH as of the vasospasm. **Conclusion:** Early suspicion, appropriate diagnosis, and a multidisciplinary approach to pregnancy-related neurological complications is crucial for achieving a successful outcome.

Keywords: complications; neurology; pregnancy; puerperium; subarachnoid hemorrhage

Sažetak. Cilj: Naglasiti važnost prepoznavanja neuroloških komplikacija u trudnoći i puerperiju. Subarahnoidalno krvarenje povezano s trudnoćom, iako nije često, dovodi do većeg morbiditeta i mortaliteta. **Prikaz slučaja:** Drugorotkinja u 39. tjednu trudnoće rodila je zdravo dijete. Neposredno prije poroda manifestirala je jaku desnostranu glavobolju praćenu zamućenim vidom. Nativni CT mozga pokazao je kortikalno subarahnoidalno krvarenje. Na CT-u, kao i na MR angiografiji mozga opisani su multifokalni vazospazmi koji su zahvatili više arterija Wilisijeva kruga te se posumljalo na sindrom reverzibilne cerebralne vazokonstrikcije (RCVS). Pacijentica je liječena nimodipinom i praćena neuroradiološki i neurosonološki u trajanju od tri mjeseca tijekom kojih dolazi do potpune resorpcije krvarenja i regresije vazospazma. **Zaključak:** Rano prepoznavanje, postavljanje točne dijagnoze i multidisciplinarni pristup neurološkim komplikacijama u trudnoći ključni su za postizanje uspješnog ishoda.

Ključne riječi: komplikacije; neurologija; puerperij; subarahnoidalno krvarenje; trudnoća

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INTRODUCTION

Convexity subarachnoid haemorrhage (cSAH) is a rare type of spontaneous, non-traumatic, non-aneurysmal SAH that occur within the cortical sulci of the brain¹⁻⁴. Since cSAH accounts for about 6% of all subarachnoid haemorrhage cases it may often go unrecognized^{1,2}. According to Cuvinciu and colleagues, cSAH is connected with different vascular and nonvascular causes, including: reversible cerebral vasoconstriction syndrome (RCVS), cerebral venous thrombosis (CVT), vascular malformations (pial arteriovenous malformations, dural arteriovenous fistulas, and cavernomas), vasculitis, infectious aneurysms, Moyamoya disease, severe atherosclerotic carotid disease, posterior reversible encephalopathy syndrome (PRES), cerebral amyloid angiopathy (CAA) and nonvascular disorders, such as primary and secondary brain neoplasms or even abscess⁵. As cSAH has various causes, it also presents with different clinical manifestations. Headache, seizures, disorientation, transient focal neurological episodes, long-term focal neurological impairments, visual abnormalities, nausea and vomiting are described by Ly et al. as typical symptoms⁶. The most common symptom, headache, manifests in two clinical forms that vary by age^{4,7}. In younger patients (≤ 60 years old) RCVS is more common followed by sudden intense headache, occasionally accompanied by neurologic impairment. Headache may be mistaken for those caused by aneurysmal SAH. On the other hand, older patients (> 60 years old) are more likely to have CAA and to present with transient sensory or motor symptoms, such as numbness, tingling, or weakness^{3,8}.

Despite being rare, pregnancy-related subarachnoid haemorrhage (pSAH) has a significant morbidity and mortality rate. Moreover, diagnosis and management of SAH in pregnancy and puerperium is challenging due to the potential harm to both the mother and foetus. As a neurological emergency, SAH requires rapid intervention and a multidisciplinary approach in order to achieve a successful outcome. We present a rare case of RCVS that occurred during the antepartum period, in contrast to the majority of cases that occur after childbirth.

CASE REPORT

41-year-old 39 weeks pregnant secundigravida was admitted to the delivery room with contractions every 3-4 minutes. During pregnancy, she was monitored closely because of gestational diabetes. The patient's medical history included migraine without aura. She did not use any licit or illicit substances (cannabis, cocaine, ecstasy amphetamines or LSD) nor any vasoactive medication (antidepressant, nasal decongestants,

We encourage readers to be aware of convexity subarachnoid haemorrhage (cSAH), a subtype of SAH that is associated with a short but significant list of underlying diagnoses that require different evaluation and treatment.

Rapid recognition of cSAH can minimise unnecessary testing and facilitate accurate prognosis and appropriate treatment.

triptans or ergot derivatives). Ten days before labour, she reported a severe pulsating headache. She subsequently gave birth to a healthy baby. Shortly before delivery, she started signalling right-sided throbbing headache and her RR was 170/100 mmHg. Because of the retained placenta, she was taken to the operation room where a manual lysis of the placenta was performed. Four hours later, she experienced confusion and reported a thunderclap right hemiparesis accompanied by blurry vision *when looking to the left that persisted for several hours*. Due to mild focal neurologic deficit, a neurologist was called and an urgent brain NCCT scan was done, which revealed a sulcal subarachnoid haemorrhage of the right frontal and parietooccipital lobe. Hunt Hess was 3, WFNS grade 1 and Fisher grade 2. Later, she reported experiencing strong pulsating headaches. The patient was transferred to the neurological intensive care unit (ICU). Subsequently, her headache disappeared, and her neurological status was normal. She was started with intravenous nimodipine therapy. CTA of the brain (Figure 1a and 1b) did not show any vascular malforma-

tions. Further on, brain MRI (Figure 1d) revealed a bilateral, but dominantly right, sulcal haemorrhage high on the convexity of the frontal lobe. There were no cavernomas, dural venous sinus thrombosis (Figure 1c), or intraparenchymal haemorrhage. MRA (Figure 1e) demonstrated

findings consistent with multifocal vasospasm (involving the left terminal ICA, M1 MCA, and A1 ACA), suggesting a case of RCVS leading to cSAH. Transcranial Doppler (TCD) revealed moderately increased flow velocities in most of the arteries (Table 1), but no vasospasm was detected. The

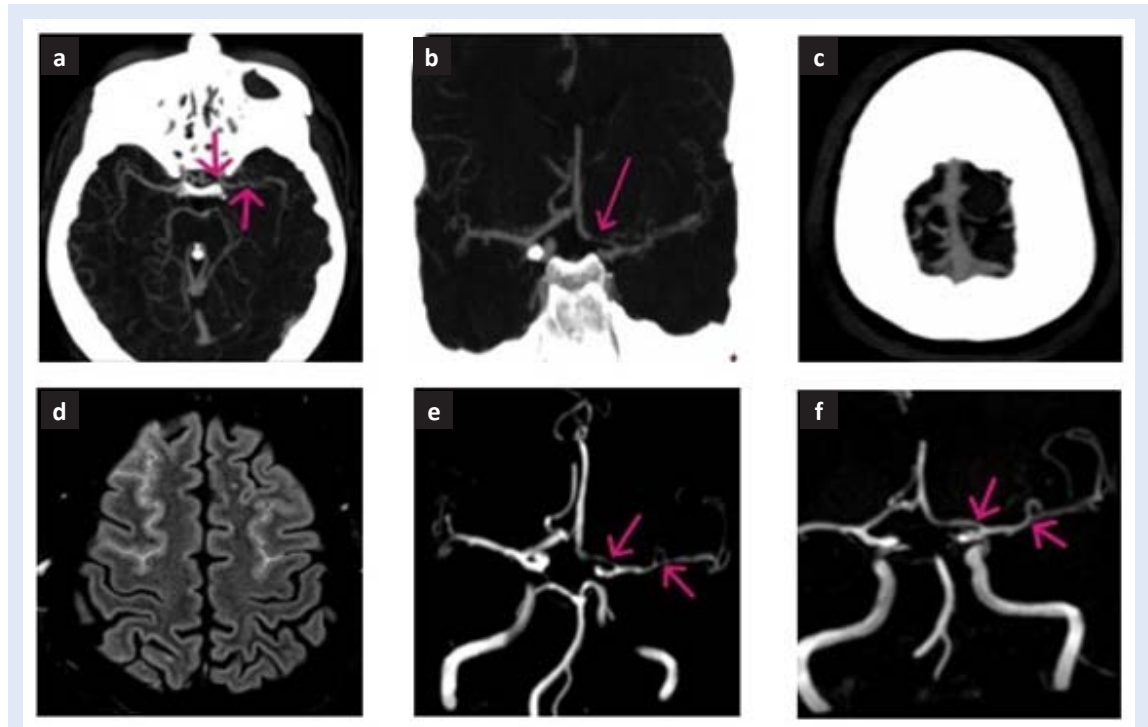


Figure 1. Reversible cerebral vasoconstriction syndrome (RCVS). CTA axial image showing vasospasm in the left ICA, M1 segment of MCA and A1 segment of ACA – arrows (a), CTA coronal image demonstrates the same vasospasm – arrows (b), CTA axial image depicting patent sagittal sinus (c), FLAIR MRI axial image showing bilateral convexity SAH (d), MRA of the circle of Willis with noticeable vasospasm – arrows indicate the most prominent changes (e), Follow-up MRA with proven resolution of vasospasm – arrows (f)

ICA, internal carotid artery; MCA, middle cerebral artery; ACA, anterior cerebral artery

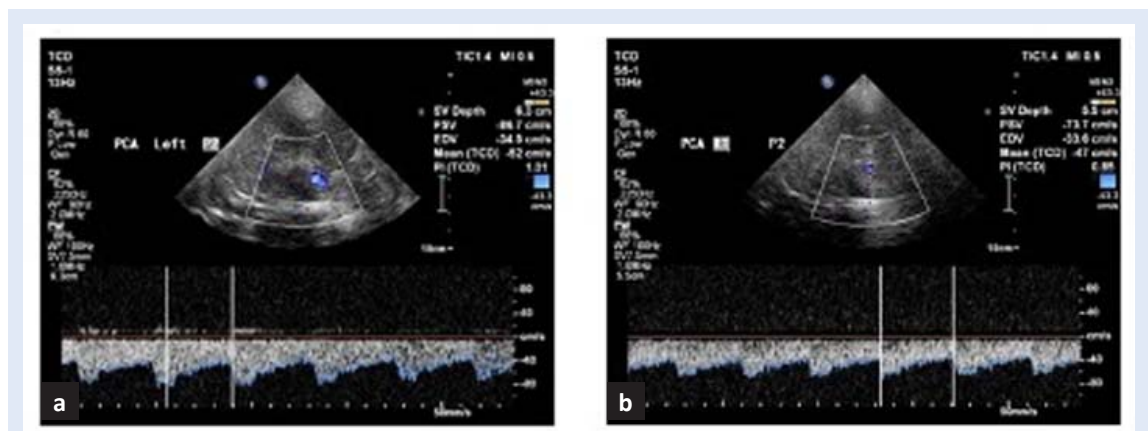


Figure 2. Transcranial color-coded sonography: P2 segment of the right PCA – increased mean flow velocity (MFV) (a), P2 segment of the left PCA – normal MFV (b)

PCA, posterior cerebral artery; MFV, mean flow velocity

Table 1. The mean flow velocity (MFV) of cerebral arteries measured from the waveform of the transcranial doppler (TCD)

Day from cSAH onset	Mean flow velocity (MFV) of cerebral arteries (cm/s)								
	R VA	L VA	BA	R MCA	L MCA	R ACA	L ACA	R PCA	L PCA
5th	35	37	55	108	104	70	71	87	63
11th	38	45	44	85	65	62	56	43	48
19th	37	40	60	81	75	60	60	58	50
46th	49	40	72	71	76	47	54	30	35
90th	45	44	46	65	65	66	59	47	52

VA, vertebral artery; BA, basilar artery; MCA, middle cerebral artery; ACA, anterior cerebral artery; PCA, posterior cerebral artery; R, right; L, left

increased mean flow velocity (MFV) was more pronounced in both right MCA and PCA despite the location of the cortical SAH (Figure 2). Control CT showed regression of the SAH. The headache no longer occurred in our patient, and she was discharged on the 12th day after the event. On discharge, a follow-up MRI was performed, which showed resolution of the SAH without new ischemic or haemorrhagic lesions, as control MRA (Figure 1f) showed a complete resolution of vasospasm. A TCD follow-up continued up to three months after the event of haemorrhage, showing blood flow normalization (Table 1).

DISCUSSION

Our case underscores the challenges clinicians encounter during management of neurological complications in pregnancy and the puerperium, as these issues can greatly impact both pregnancy and childbirth, with potential for various short- and long-term sequelae.

SAH, as emergency requires immediate treatment and it is particularly concerning in the context of pregnancy due to pregnancy-related modifications, particularly those impacting hemodynamic stability and cerebral autoregulation that need to be considered during treatment⁹.

In pregnancy, physiological alterations such as modified cardiovascular hemodynamic, adjusted coagulation profiles, and hormonal fluctuations are believed to contribute to the development of SAH¹⁰. It is controversial if pregnancy raises the risk of SAH and whether the risk is higher in specific stages of pregnancy or puerperium¹¹.

In a nationwide analysis from the United States, the incidence of pregnancy-related pSAH was 5.8–8.5/100 000 deliveries^{9,12}. In pregnancy, ac-

Our case highlights the importance of a comprehensive evaluation of a woman experiencing a sudden headache during pregnancy or within the first six weeks following childbirth.

ording to a Finnish population-based study, aneurysmal subarachnoid haemorrhage (aSAH) is more frequent than non-aneurysmal SAH (non-aSAH); respectively the incidence was 2,48 compared to 0,73 per 100 000 deliveries¹¹. aSAH was also related to older patients (40 years of age or more) and with a statistically greater possibility for rupturing in the third trimester¹¹. Better outcomes were confirmed during the second trimester, when the incidence of a non-aSAH peaked¹³. As previously mentioned, aetiology of cSAH varies according to patient age: in patients under 60 years, RCVS is the most frequent cause, while CAA is more prevalent in individuals over 60¹⁴. Regarding pregnancy-related cSAH, potential mechanisms to investigate include RCVS, as in our case. Other mechanisms, such as PRES and CVT were ruled out in our patient after comprehensive evaluation. In our case the patient suffered cSAH secondary to RCVS.

RCVS is described clinically by the sudden onset of severe headache, sometimes accompanied with neurologic deficits, haemorrhage or ischemia⁷. In our patient, cSAH manifested as a thunderclap headache along with visual disturbances, with RCVS confirmed to be associated with pregnancy and a history of migraines.

In RCVS, CT or MR angiography identifies diffuse and segmental narrowing or "beading" of multiple intracranial arteries, suggesting vasospasm, usually in multiple vascular territories^{7,15}. TCD is

used to non-invasively *monitor* the development and temporal *course* of cerebral *vasospasm*^{7,16}. In the case we presented, the blood flow through the extracranial carotid and vertebral arteries was normal, as determined by *Colour Doppler (CD)* and *TCD analysis*. Initial transcranial color-coded Doppler (TCCD) showed moderately increased flow velocities in most of the arteries, more evident in both right MCA and PCA, that can be linked to a compensatory mechanism. Those values, though, were not consistent with a definition of vasospasm. That increased velocity tended to dissolve on our next follow-up, on the 11th day. The difference between angiography and TCCD findings can be attributed to the reduced sensitivity of TCD, which is between 42 % to 67 % compared to the 80 % of a CTA or MRA and 100 % for a DSA¹⁵. The increase of flow velocities in TCD are not as severe as in aneurysmal SAH but its presence in P2 shows a higher risk of developing PRES or an infarct in the M1 segment¹⁷. Spatial and temporal spread of vasospasm initially affects very small, distal arteries and later medium and large-sized arteries¹⁸. Such involvement might explain often early normal angiogram and the absence of typical TCD vasospasm. With the complete vasospasm resolution within 3 months, the last criteria for RCVS were met in our patient.

Diagnostic criteria for RCVS are⁷:

- thunderclap headache with or without focal deficit or seizures
- monophasic course
- segmental vasoconstriction of cerebral arteries
- no evidence of aneurysmal SAH
- normal CSF
- complete normalization of arteries within 12 weeks.

Reports indicate that potential triggers for RCVS include antimigraine agents: triptans and ergotamines, decongestant medications such as pseudoephedrine, diet pills and energy-enhancing agents with amphetamine derivatives such as ephedrine, and illicit drugs such as cocaine and amphetamine¹⁹. Pregnancy is the most common factor contributing to RCVS, with more than half of all RCVS cases occurring after childbirth (postpartum), especially within the first six weeks^{7,20}. Other pregnancy related triggers of RCVS include

epidural anaesthesia, postpartal haemorrhage or inhibition of lactation²⁰.

As previously mentioned, RCVS is more frequently observed in postpartum than pregnant women, and it was once known as postpartum angiopathy²¹. In our case, RCVS manifested during the antepartum period, which makes our case rare.

The management of cSAH includes treating complications, removing potential triggers, minimising the risk of haemorrhage extension and recurrence, and addressing underlying causes. Calcium channel blockers, including nimodipine and verapamil are commonly used in RCVS^{2,22}. Results from two prospective case series indicate that nimodipine does not influence the temporal progression of cerebral vasoconstriction, but it may decrease both the occurrence and intensity of headaches. Additionally, nimodipine has been shown to have an impact on smaller blood vessels that are not easily detectable through angiography. If utilized, calcium channel blockers may be discontinued once symptoms or angiographic abnormalities have resolved^{23,24}. Regarding RCVS, the significance of recognizing and avoiding known triggers cannot be overstated.

The prognosis of cSAH is highly dependent on the underlying causes. RCVS generally tends to have a good prognosis^{2,14}. As in our patient, RCVS is considered a benign, monophasic disease with complete resolution of symptoms within weeks and vasospasm after 3 months¹⁶. However, about 1,4 % of cases had a fatal outcome. Fugate et al. reported a series of 4 fulminant postpartal cases of RCVS, with cerebral oedema, ischemia or intracranial bleeding²⁵.

CONCLUSION

Neurological complications in pregnancy and the puerperium pose a challenging task for clinicians and they deserve particular attention due to varied symptomatology and risk of morbidity and mortality to the mother and the foetus. Hence, a multidisciplinary approach is mandated to determine prompt and accurate diagnosis and direct the safest therapeutic path for the patient and the unborn child.

Conflicts of Interest: Authors declare no conflicts of interest.

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