

ISCHEMIC STROKE DUE TO POSTPARTUM ANGIOPATHY COMPLICATED BY PULMONARY EMBOLISM WITH FAVORABLE OUTCOME

Małgorzata Wiszniewska and Amelia Bytowska

Department of Neurology, Piła Specialist Hospital, Piła, Poland

SUMMARY – Postpartum cerebral angiopathy is a relatively rare condition. It can cause either ischemic or hemorrhagic stroke, or both, and usually occurs within the first week following non-complicated pregnancy and natural delivery. Although its pathophysiology is unclear, the cause of the condition is believed to be prolonged reversible vasospasm. We present an unusual case of a 37-year-old woman who developed right hemiparesis with aphasia on day 8 of natural delivery complicated by pulmonary embolism. Steroids, heparin, and calcium channel blockers were successfully instituted and the patient was discharged from the hospital on day 50. The article presents clinical and imaging characteristics, differential diagnosis, management, and considers the difficulties that occurred during the patient's hospital stay.

Key words: *Postpartum angiopathy; Cerebral vasospasm; Headache; Ischemic stroke*

Introduction

Postpartum angiopathy (PPA), a reversible segmental cerebral vasoconstriction syndrome (RCV), is a clinical-angiographic medical condition characterized by an abrupt onset of thunderclap headaches, seizures, focal neurological deficits, segmental narrowing and dilatation of large and medium-sized cerebral arteries within several days of natural delivery. Sometimes it can be complicated by ischemic stroke¹⁻⁷. Computed tomography angiography or magnetic resonance angiography (CTA, MRA) shows segmental vasoconstriction in the arteries leading to the brain and in the cerebral arteries^{1,2,7-10}. In treating the condition, calcium channel blockers, intravenous magnesium, steroids and heparin have been used, with variable success.

Correspondence to: *Małgorzata Wiszniewska, MD, PhD*, Piła Specialist Hospital, Rydygiera 1, 64-920 Piła, Poland
E-mail: mpwysz@gmail.com

Received February 20, 2012, accepted December 15, 2012

Case Report

On day 8 following natural and uncomplicated delivery, a 37-year-old woman developed a sudden-onset severe occipital headache and right hemiparesis. On admission to neurological department, she could not walk and had right hemiparesis with aphasia and hemi-right blindness. Brain imaging findings included multiple ischemic lesions in the posterior border-zone regions in the left cerebral hemisphere (CT), a diffuse cerebral and extracranial segmental arterial constriction (CTA) (Fig. 1), and multiple T2-weighted ischemic lesions in the left hemisphere of the brain and cerebellum (MRI). Erythrocyte sedimentation rate was elevated (50 mm/h), but extensive tests for vasculitis and coagulopathy were negative.

The treatment consisted initially of intravenous magnesium, saline, compound electrolyte solution, subcutaneous 0.4 mL nadroparin, and steroids. However, on day 26, dyspnea occurred. D-dimers were elevated and echocardiography revealed a thrombus in the pulmonary trunk. After pulmonary

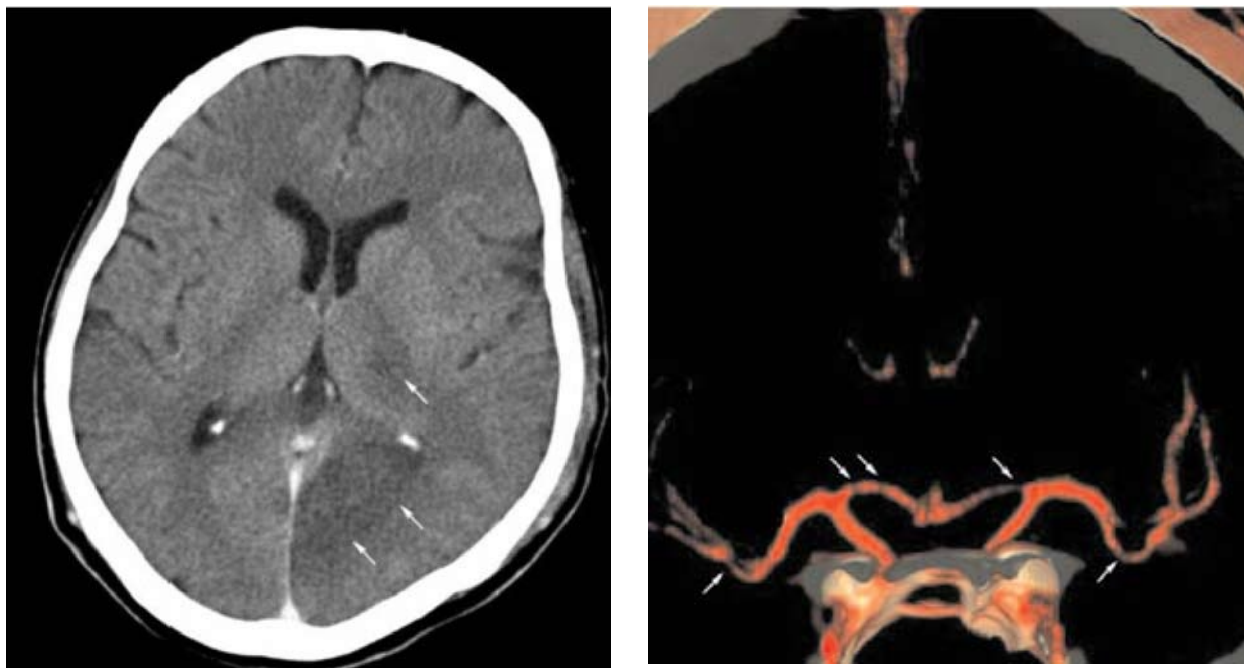


Fig. 1. Multiple ischemic lesions in posterior border zone regions of the left cerebral hemisphere with the space-occupying lesions of the left occipital cortex and multifocal vasoconstrictions in the extracranial and intracranial arteries (white arrows).

embolism had been recognized, a full therapeutic dose of nadroparin was administered; nevertheless, the patient's condition did not improve. Therefore, nadroparin was discontinued and unfractionated heparin was instituted intravenously under activated partial thromboplastin time control. After 14 days, pulmonary embolism receded, and oral warfarin treatment was started. The patient's clinical status improved gradually, and she was discharged from the hospital on day 50 with 3 points on the modified Rankin scale (mRS). Two years after the event, the patient felt well (mRS 2 points). Follow up CT and MRI showed only old lesions in the brain.

Discussion

The condition manifested clinically in the postpartum period. Brain CT and MRI showed posterior-predominant ischemic lesions, while CTA demonstrated segmental arterial vasoconstriction and narrowing. Extensive tests for cerebral vasculitis and thrombophilia were negative. There was no vascular risk factor. Both clinical and imaging features in the patient sug-

gested PPA^{1,7,11-13}. Typically for RCV^{2,3,8-14}, the course of the disease was fluctuating. What was interesting and unusual in this particular case was the fact that RCV coincided with pulmonary embolism. A similar case has not been reported in the literature so far.

To minimize the risk of blood loss during delivery, normal pregnancy is associated with a manifest shift towards hypercoagulability in the coagulative and fibrinolytic systems. However, this also increases the risk of thromboembolism. Moreover, according to the literature, in the postpartum period, dysfunction of endothelium in vessels may occur^{1,8,9,15}, and this mechanism might have led to the development of pulmonary embolism in our patient. Prompt and determined treatment resulted in favorable outcome: pulmonary embolism regressed completely and cerebral symptoms were considerably diminished. This shows that in the given circumstances, it is appropriate to convert from low molecular weight heparin to unfractionated heparin. This also proves that the coexistence of PPA and pulmonary embolism is possible; yet, the condition can be successfully treated.

References

1. SINGHAL AB, BERNSTEIN RA. Postpartum angiopathy and other cerebral vasoconstriction syndromes. *Neurocrit Care* 2005;3:91-7.
2. SATTAR A, MANOUSAKIS G, JENSEN M. Systematic review of reversible cerebral vasoconstriction syndrome. *Expert Rev Cardiovasc Ther* 2010;8:1417-21.
3. SINGHAL AB. Cerebral vasoconstriction syndromes. *Top Stroke Rehabil* 2004;11:1-6.
4. MISIRLI H, DOMAC FM, YILDIZ O. Acute cerebrovascular diseases during pregnancy and postpartum period. *Turk Serebrovaskuler Hastaliklar Dergisi* 2008;14:15-8.
5. KUKLINA EV, TONG X, BAUSIL P, GEOGARGE MC, CALLAGHAN WM. Trends in pregnancy hospitalizations including a stroke in the United States from 1994 to 2007: reason for concern. *Stroke* 2011;42:2564-70.
6. Del ZOTTO E, GIOSSI A, VOLONGHI I, COSTA P, *et al.* Ischemic stroke during pregnancy and puerperium. *Stroke Res Treat* 2011;606780. Doi: 4061/2011780. Published online.
7. IVANKOVIĆ M, BOGOJE-RASPOPOVIĆ A, DROBAC M, MAMIĆ-MARTINOVIĆ D, VODOPIĆ M. Benign angiopathy of the central nervous system or reversible cerebral vasoconstriction syndrome. *Acta Clin Croat* 2011;50:253-5.
8. SINGHAL AB. Postpartum angiopathy with reversible posterior leukoencephalopathy. *Arch Neurol* 2004;61:411-6.
9. CALABRESE LH, DODICK DW, SCHWEDT TJ, SINGHAL AB. Narrative review: reversible cerebral vasoconstriction syndromes. *Ann Intern Med* 2007;146: 34-44.
10. LAKHDAR R, BAFFOUN N, HAMMAMI N, NAGI S, BACCAR K, DRISSI S, KADDOUR C. Neuroradiological pattern of peripartum cerebrovascular disease medicating transfer to determine care unit. *Tunis Med* 2012;90:223-32.
11. FUGATE JE, AMERISO SF, ORTIZ G, SCHOTTLAENDER LV, WIJDICK EF, FLEMMING KD, RABINSTEIN AA. Variable presentations of postpartum angiopathy. *Stroke* 2012;43:670-6.
12. TATE J, BUSHNELL C. Pregnancy and stroke risk in women. *Womens Health* 2011;7:363-74.
13. LEMMENS R, SMET S, WILMS G, DEMAEREL P, THIJS V. Postpartum RCVS and PRES with normal initial imaging findings. *Acta Neurol Belg* 2012;112:189-92.
14. MISIRLI H, MAYDA DOMAC F, YILDIZ O. Acute cerebrovascular disease during pregnancy and postpartum period. *J Turk Cerebrovasc Dis* 2008;14:15-8.
15. KATZIN LW, LEVINE M, SINGHAL AB. Dural puncture headache, postpartum angiopathy, pre-eclampsia and cortical vein thrombosis after an uncomplicated pregnancy. *Cephalgia* 2009;29:791-5.

Sažetak

ISHEMIJSKI MOŽDANI UDAR UZROKOVAN POSTPARTALNOM ANGIOPATIJOM I PLUĆNOM EMBOLIJOM S POVOLJNIM ISHODOM

M. Wiszniewska i A. Bytowska

Postpartalna cerebralna angiopatija je relativno rijetko stanje koje, međutim, može uzrokovati ishemijski ili hemoragijski moždani udar ili pak oba, a obično nastaje u prvom tjednu nakon trudnoće bez komplikacija i prirodnog porođaja. Iako patofiziologija ovoga stanja nije razjašnjena, smatra se da ga uzrokuje produženi reverzibilni vazospazam. Prikazujemo neuobičajeni slučaj 37-godišnje žene kod koje se razvila desnostrana hemipareza s afazijom 8. dana nakon prirodnog porođaja s komplikacijom plućne embolije. Uvođenje steroida, heparina i blokatora kalcijevih kanala pokazalo se uspješnim i bolesnica je otpuštena iz bolnice 50. dana bolničkog liječenja. Prikazuju se kliničke i slikovne značajke, diferencijalna dijagnostika i liječenje, uz osvrt na teškoće koje su se javile tijekom bolesničina boravka u bolnici.

Ključne riječi: *Postpartalna angiopatija; Cerebralni vazospazam; Glavobolja; Ishemijski moždani udar*

