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Case report

# "A case of postpartum headache post dural puncture. Post Dural Puncture Headache or Reversible Cerebral Vasoconstriction Syndrome - Posterior Reversible Encephalopathy Syndrome?"

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**Summary.** *Background and aim:* Reversible Cerebral Vasoconstriction Syndrome (RCVS) and Posterior Reversible Encephalopathy Syndrome (PRES) are two rare neurological conditions, clinically characterized by headache. In our case a diagnosis of PDPH was made though imaging showed signs of RCVS-PRES. *Methods:* We present a case of RCVS-PRES in a postpartum woman who presented headache as first symptom and only later experienced seizures. Dural puncture worked as a confounding factor in the clinical postpartum evaluation. *Results-Conclusions:* We want to focus the attention on changes of clinical characteristics of headache as an important factor to be analysed, in order to have a prompt diagnosis. We therefore propose a diagnostic algorithm. Moreover, we evaluate possible triggers of RCVS and PRES; in our case dural puncture is probably not the trigger, in fact there were no liquoral hypotension signs on imaging.

**Key words:** Post Dural Puncture Headache, Posterior Reversible Encephalopathy, Postpartum, Reversible Cerebral Vasoconstriction Syndrome

## Background

Reversible Cerebral Vasoconstriction Syndrome (RCVS) is a rare condition characterized by thunderclap headache and reversible vasoconstriction of the cerebral arteries. It is a cerebrovascular disorder caused by a transient disregulation of cerebral vascular tone, leading to multifocal arterial constriction and dilation. The primary clinical manifestation is recurrent sudden-onset and severe headache over 1-3 weeks, often accompanied by nausea, vomiting, photophobia, confusion and blurred vision. Posterior Reversible Encephalopathy Syndrome (PRES) is a syndrome characterized by reversible posterior brain vasogenic edema, most commonly in the parieto-occipital regions. It's caused by a rapid increase of arterial blood pressure that can lead to cerebral hyperperfusion with consequent vascular leakage and vasogenic edema. It seems to be triggered by sympathetic hyperactivity and endothelial dysfunction caused by circulating endogenous or exogenous substances. Neurological symptoms, including headache, visual deficits, disorders of consciousness, confusion, seizures and focal neurological signs are appreciated in this condition as well. These two syndromes share endothelial dysfunction as a pathophysiological feature (1,2). Scientific literature about coexistence of PRES-RCVS-post dural puncture headache (PDPH) describes cases of women who had a history of migraine, preeclampsia or in good clinical conditions that underwent spinal or epi-

dural anesthesia with inadvertent dural puncture (3-7). They presented a severe headache between day 2 and 7 after the procedure, and alteration of the consciousness status between day 3 and 7. The diagnosis was reached only after imaging (8, 9). Soon afther that, medication that comprehend antiepileptic drugs (Levetiracetam, Phenytoin, $MgSO_4$) and Nimodipin for the vasospasm was started. All these papers try to find a correlation between dural puncture and RCVS-PRES, and they all agree in suggesting that dural puncture could be a possible trigger in a substrate of disorders of the cerebral vascular function.

We present a case of RCVS-PRES in a postpartum woman with inadvertent dural puncture during epidural analgesia catheter positioning. We point the attention on clinical characteristics and timing of the headache as fundamental data to have a prompt diagnosis and treatment. Then we consider the possibility for dural puncture to be a trigger for RCVS-PRES.

## Case

A 31 years old woman had an induced labour for fetal macrosomia. A spinal-epidural analgesia was performed. Epidural catheter positioning was difficult and suspected for dural puncture. After 12 hours of labour analgesia C-delivery (Cesarean-delivery) was performed because of labour dystocia. No surgical or anesthesiological complications were reported.

24 hours after dural puncture the patient developed bilateral and orthostatic headache with no associated symptoms so PDPH was diagnosed, and therapy with caffeine, paracetamol, bed rest and hydration was prescribed. Progressive improvements of symptoms were observed. On day 4 she developed a severe, frontal, non postural headache associated with nausea, dizziness, tinnitus and no longer responsive to PDPH therapy. On day 5 alterations of the consciousness appeared with 3 episodes of psychomotor agitation followed by 2 tonic-clonic seizures. At the end of the episodes neurological examination showed isochoric and isocyclic pupils, no trouble speaking, no numbness or weakness on one side of the body, no other signs of stroke. No hypertension and proteinuria were detected so preeclampsia was excluded.

A Computed tomography scan (CT scan) showed focal hyperdensity in the basal occipital right area (Fig.1)

The Magnetic Resonance Imaging (MRI) revealed signs of vasogenic edema in the bilateral hemispheres (Fig.2).

Magnetic Resonance Angiography (MRA) showed segmental vasospasm with size reduction of P3 segment of left posterior cerebral artery, P4 bilateral, M1 segment of medium cerebral arteries (Fig. 3).

No signs of liquoral hypotension were recorded. Imaging findings were consistent with both RCVS and PRES. The patient was transferred to Intensive Care Unit (ICU) and was evaluated by the neurologist who set up a therapy with Lacosamide and Nimodipine. Electroencephalogram (EEG) showed moderate slow activity in the right posterior areas, mild on the left, and no signs of comitial crises. After 2 days no cephalea or other symptoms were recorded and the patient was transferred to the postpartum ward. On day 14 the MRI showed resolution of the previous findings (Fig.4) and the spinal MRI was negative for signs of leak.

The patient was then discharged from hospital in good clinical conditions.

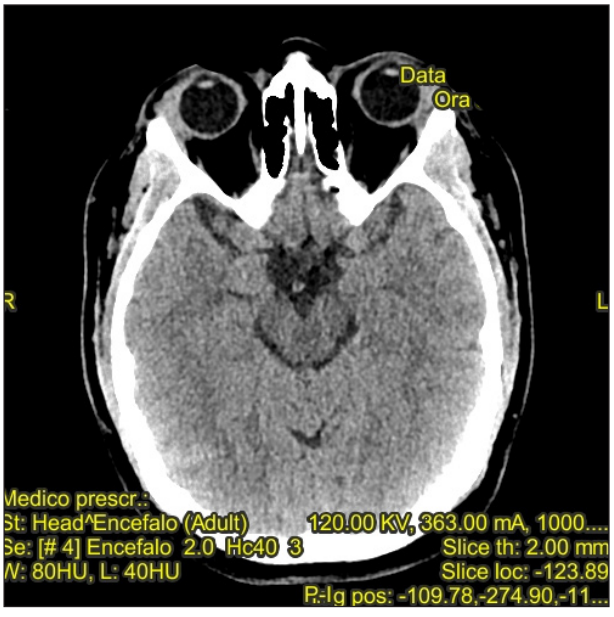


**Figure 1.**

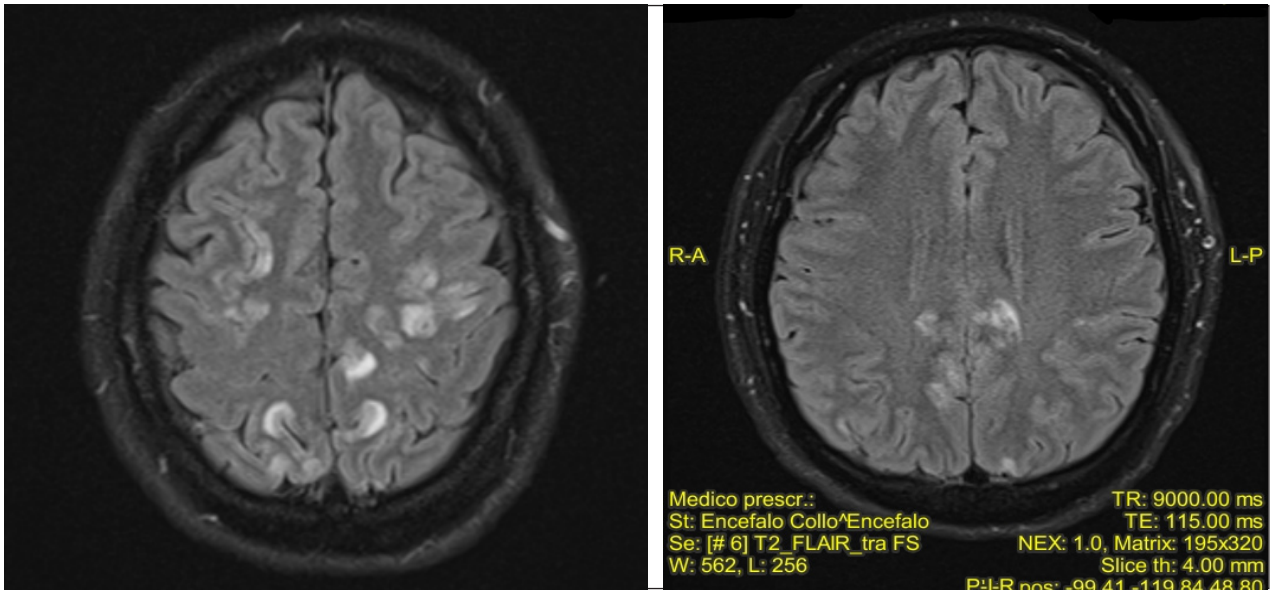


**Figure 2.**

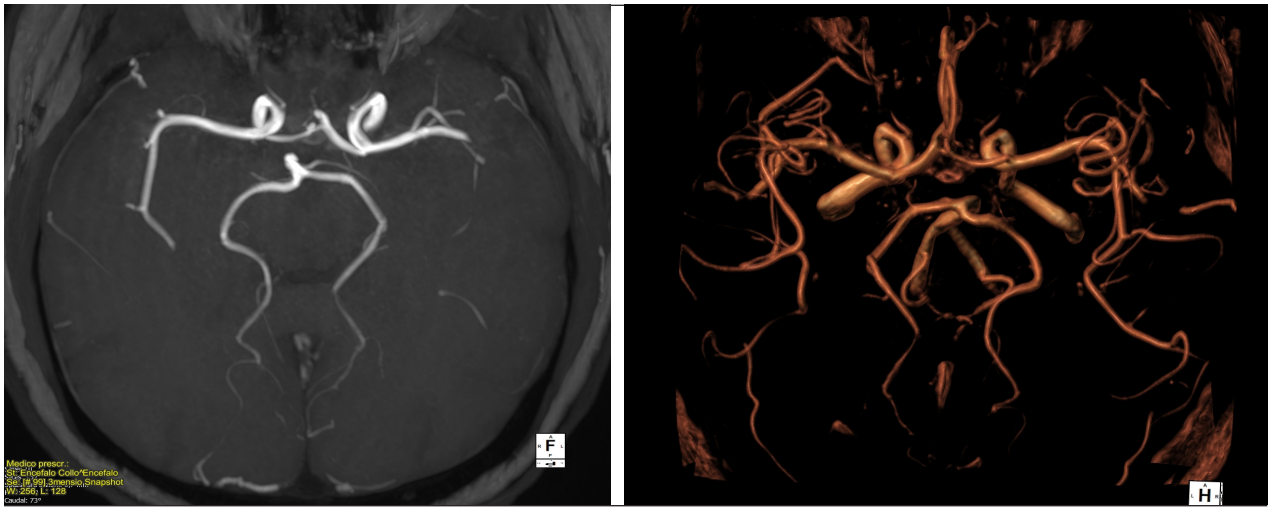

**Figure 3.**

## Conclusion

Due to the different possible aetiologies of postpartum headache a careful evaluation of clinical characteristics of symptoms is crucial. In our case clinical features of headache changed during postpartum. On day 1 to 3 it was a postural, bilateral persistent headache with no associated symptoms so, according to the International Classification of Headache Disorders criteria, it was a PDPH (10). A therapy with bed rest, hydration, caffeine and paracetamol was prescribed with a good clinical result. On day 4 the situation changed as the headache was severe, no longer positional, associated with tinnitus, dizziness and no longer responsive to PDPH therapy. These clinical characteristics could have led us to get immediately CT scan or preferably MRI, before appearance of seizures. Once diagnosed, it was clear that headache was

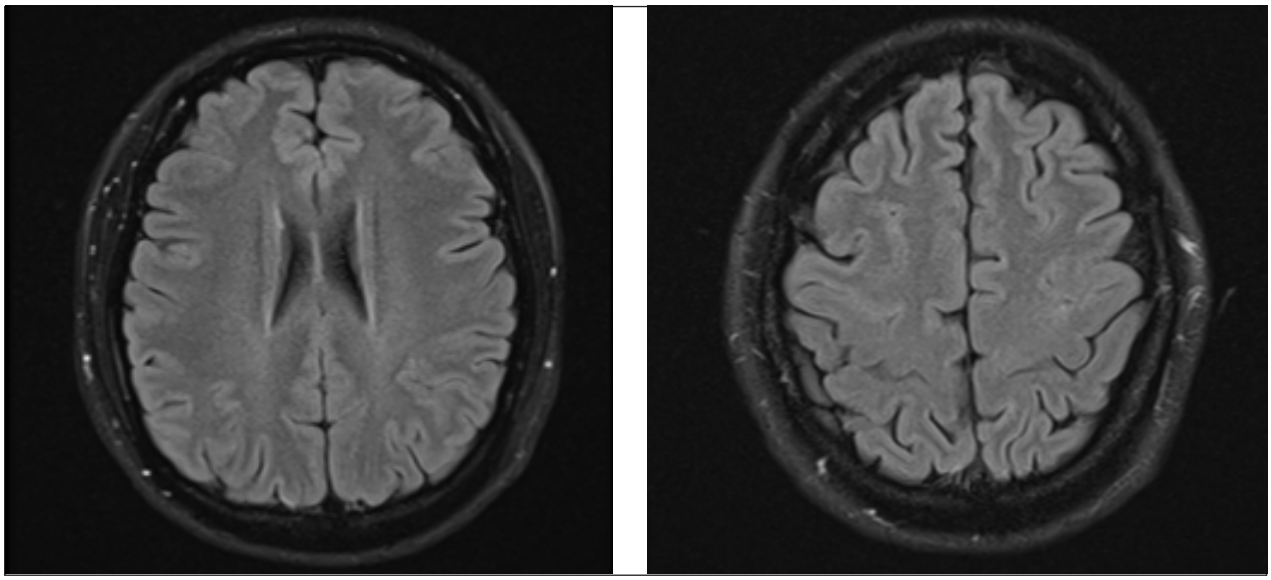

**Figure 4.**

typical of RCVS-PRES. History of dural puncture was a confounding factor that delayed diagnosis for a few hours even if final result was however favorable.

This case wants to focus attention on the importance of an accurate evaluation of headache characteristics and its changes, in order to have a prompt diagnosis and start an appropriate therapy in time. We therefore propose a diagnostic algorithm. If postpartum headache lasts for more than 24 hours and neurological changes appear: 1)define clinical characteristics of headache according to International Classification of Headache Disorders 2)measure blood pressure 3) perform blood tests 4)perform urinary tests. Once diagnosis of PDPH, preeclampsia and primary headache disorders are excluded immediate imaging is indicated.

The second point we want to stress is if dural puncture can be considered a trigger for RCVS-PRES. As previously discussed, these syndromes might be triggered by circulating endogenous or exogenous factors leading to endothelial activation.

Puerperium itself can be a condition that promotes endothelial dysfunction in a substrate of disorders of the cerebral vascular function. In our case, differently from cases reported in literature, there was no evidence of liquoral hypotension on imaging. The relationship between dural puncture and RCVS-PRES is therefore inconsistent. Radiologic findings show a typical RCVS-PRES presentation (arterial vasoconstriction typical for RCVS and edema of the posterior areas characteristic of PRES) in absence of liquoral hypotension signs or lumbar alterations in the puncture site.

**Conflict of Interest:** Each author declares that he or she has no commercial associations (e.g. consultancies, stock ownership, equity interest, patent/licensing arrangement etc.) that might pose a conflict of interest in connection with the submitted article

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