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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Case report

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Abstract

BACKGROUND

Reversible Cerebral Vasoconstriction Syndrome (RCVS) and Posterior Reversible Encephalopathy Syndrome (PRES) are two rare neurological conditions, clinically characterized of headache. This is sometimes difficult to differentiate from post dural puncture headache (PDPH). In our case a diagnosis of PDPH was made but imaging showed signs of RCVS-PRES. The novelty is that, unlike in cases reported in literature, no liquoral hypotension signs were detected on imaging.

CASE PRESENTATION

We present a case of RCVS-PRES in a postpartum woman that presented headache as first symptom, and only later experienced seizures. Epidural analgesia was performed during labour, and it was complicated by dural puncture that worked as a confounding factor in the clinical postpartum evaluation. Seizures represented an unexpected event in the course of treating this patient for post dural puncture headache.

CONCLUSIONS

We point out the attention on changes of clinical characteristics of headache as an important factor to be analyzed, in order to have a prompt diagnosis. 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. According to literature reports, puerperium itself was a promoting factor.

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. Most cases mainly occur during postpartum period or after exposure to vasoactive substances. 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. The major complications are subarachnoid hemorrhage and stroke. Incidence is unknown but it does not appear rare on the basis of prospective and retrospective studies.

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 disfunction caused by circulating endogenous or exogenous substances that might be responsible of endothelial activation resulting in release of immunogenic and vasoactive substances. Neurological symptoms, including headache, visual deficits, disorders of consciousness, confusion, seizures and focal
neurological signs are appreciated in this condition as well. The incidence of PRES in pregnancy or puerperium is not known, eventhough this syndrome is increasingly diagnosed.\textsuperscript{4,5}

These two syndromes share endothelial dysfunction as a pathophysiological feature.\textsuperscript{6,7}

Scientific literature about coexistence of PRES-RCVS-post dural puncture headache (PDPH) is poor and limited to case reports.\textsuperscript{8–12} This reports describe cases of women who had a history of migraine, preeclampsia or in good clinical conditions that underwent Cesarean delivery (C-delivery) with spinal or epidural anesthesia with inadvertent dural puncture. They presented a severe headache between day 2 and 7 after Cesarean section (C-section), and alteration of the consciousness status between day 3 and 7. The diagnosis was reached only after imaging\textsuperscript{13–15}. Soon after that, medication that comprehend antiepileptic drugs (Levetiracetam, Phenytoin, MgSO\textsubscript{4}) and Nimodipin for the vasospasm, was started. All this 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.

\textbf{Case Presentation}

A 31 years old woman had an induced labour for fetal macrosomia at gestational age 37 + 4. A combined spinal epidural analgesia was performed with spinal access L4-L5 while epidural catheter positioning was difficult and suspected for dural puncture (level L3-L4). Aspiration test was positive for cold and clear liquid, test dose with lidocaine 1\% 3 ml was negative for motor block and uncertain for sensitive block because of poor woman collaboration in describing sensitivity. Top up technique was performed, and it requested bolus additions that reached epidural dosages to have an adequate analgesia. After 12 hours of labor analgesia C-delivery was performed because of labor dystocia. Both epidural and spinal anesthesia were unsuccessfull so general anesthesia was performed. 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 speakig, 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. Arterial vasoconstriction was typical for RCVS and edema of the posterior areas was characteristic of PRES. The patient was transferred to Intensive Care Unit (ICU) and was evaluated by the neurologist that 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 cefalea 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.

Discussion And Conclusions

Due to the different possible etiologies of postpartum headache a careful evaluation of clinical characteristics of symptoms is crucial.\textsuperscript{16,17} 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.\textsuperscript{18} A therapy with bed rest, hydration, caffeine and paracetamol was prescribed with a good clinical result.\textsuperscript{19} 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. This clinical characteristics should have led us to get immediately CT scan or preferably MRI, before appearance of seizures. Once diagnosed, it was clear that headache was 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.

The second point we want to analyse is if dural puncture can be considered a trigger for RCVS-PRES. Literature, about dural puncture as a possible trigger, is poor and unconsistent.\textsuperscript{8–12} 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.

**Abbreviations**

RCVS
Reversible Cerebral Vasoconstriction Syndrome
PRES
Posterior Reversible Encephalopathy Syndrome
PDPH
post dural puncture headache
C-delivery
Cesarean delivery
C-section
Cesarea section
CT scan
Computed tomography scan
MRI
Magnetic Resonance Imaging
MRA
Magnetic Resonance Angiography
ICU
Intensive Care Unit
EEG
Electroencephalogram

**Declarations**

**Ethics approval and consent to partecipate:** Yes

**Consent for publication:** Written informed consent was obtained from the patient for publication of this case report and accompanying images

**Availability of data and material:** data sharing not applicable to this article as no datasets were generated or analysed during the current study

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21. DECLARATIONS

Figures

![CT scan: signs of focal hyperdensity in the basal occipital right area](image)

**Figure 1**

CT scan: signs of focal hyperdensity in the basal occipital right area
Figure 2

MRI: signs of vasogenic edema in the bilateral hemispheres

Figure 3

MRA: segmental vasospasm with size reduction of P3 segment of left posterior cerebral artery, P4 bilateral, M1 segment of medium cerebral arteries
Figure 4

Resolution of vasogenic edema signs.