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Case Report

Late postpartum eclampsia: A time-critical diagnosis

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Abstract

Eclampsia is known to cause posterior reversible encephalopathy syndrome (PRES), which is commonly associated with extensive neurovascular damage affecting preferably posterior regions and often leads to reversible cortical blindness. We describe a case of a young woman with PRES in a late postpartum stage. The patient was not initially diagnosed with preeclampsia nor PRES due to the presence of ambiguous symptoms. A diagnosis of PRES and late postpartum eclampsia (LPE) was obtained by eliminating other more likely diagnoses consistent with the confounding symptoms. Finally, once appropriate treatment was instituted, the patient's PRES was resolved without neurological sequelae. This case emphasizes the importance of properly and quickly recognizing PRES and LPE, as tardy or inappropriate treatment can lead to irreversible neurological damage.

Key Words:

Late postpartum eclampsia; posterior reversible encephalopathy syndrome; postdural puncture headache; cortical blindness; seizure

Introduction

The reported case occurred in a tertiary level multidisciplinary center. The in-hospital medical emergency team was activated for a patient with seizures and cortical visual loss in the late postpartum period. In the preceding hours, the patient showed signs of hypertensive emergency which were dismissed due to the existence of a complication from epidural anesthesia, and also because physicians were unaware that, although rare, eclampsia may occur more than 48 hours postpartum. This case report illustrates the difficulties that are frequently encountered in recognizing and diagnosing LPE, and underlines the importance of evaluating postpartum patients carefully for signs and symptoms suggestive of LPE.

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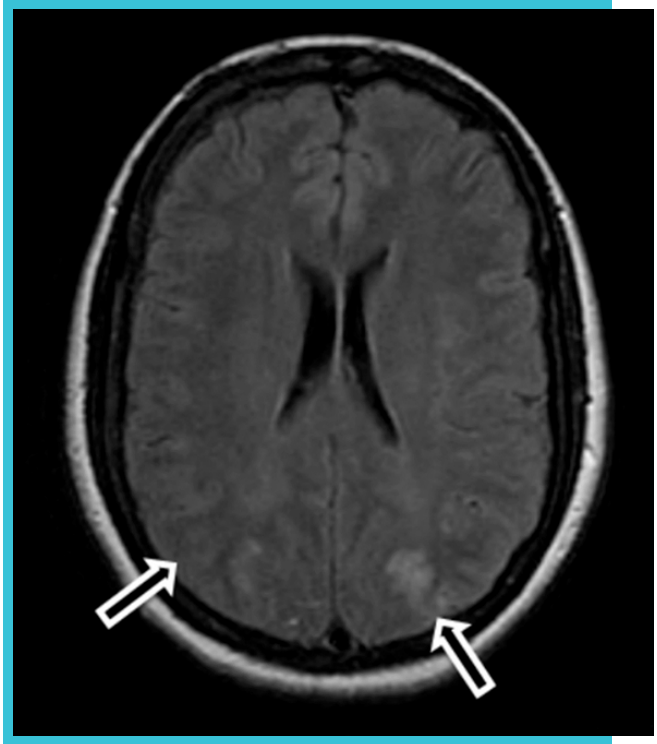
Case Presentation

A 26-year-old healthy woman, gravida 2 para 1, was admitted at 41 weeks of gestation for labor induction owing to post term pregnancy and fetal macrosomia. Patient medical and obstetric history, including her previous pregnancy 2 years earlier, was unremarkable and neither proteinuria nor arterial hypertension was detected. The patient's family history was also unremarkable. An epidural catheter was placed for labor analgesia. As a result of failed block, a second epidural catheter was introduced with satisfactory sensorial analgesia without motor block. Aspiration through the catheters was negative for cerebrospinal fluid (CSF) or blood, and no other complications were described. During labor, the patient's blood pressure

(BP) remained within normal limits (patient baseline 120/65 mmHg) and a urine test strip was negative for proteins. Due to fetal distress, the patient underwent an emergent caesarean section under spinal anesthesia. Cardiorespiratory parameters remained stable throughout the surgery.

Figure 1.



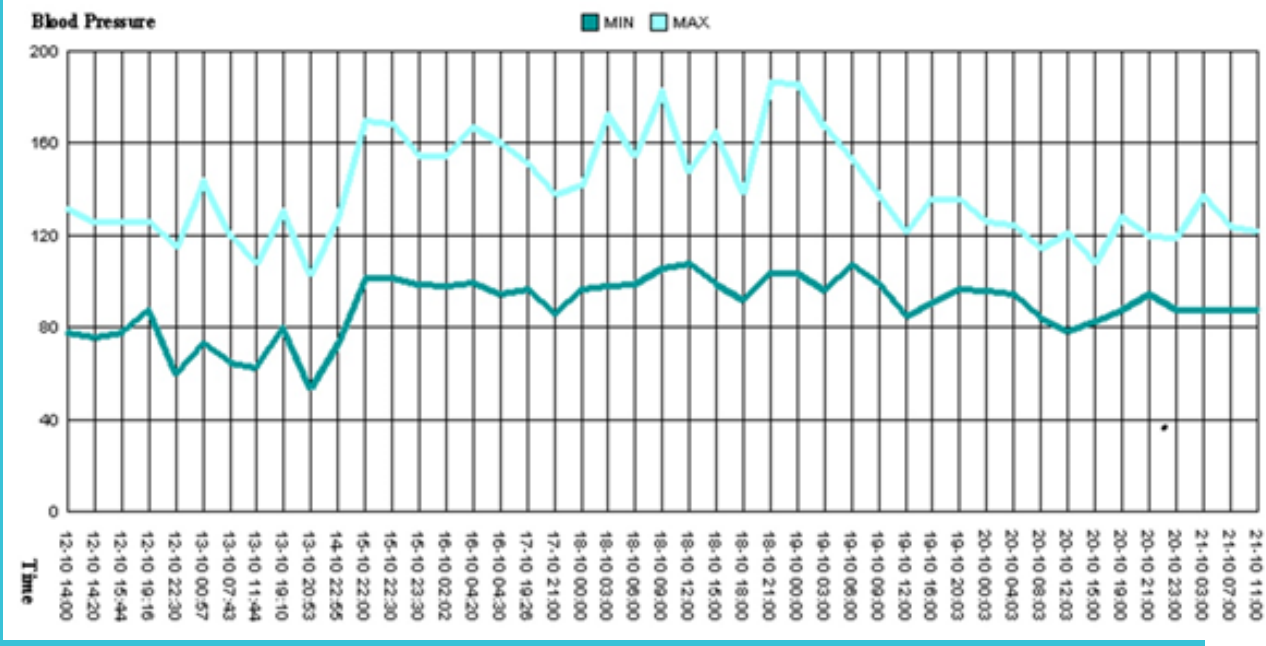
T2-Weighted magnetic resonance image obtained following the patient's seizures. The image shows bilateral parieto-occipital hyperintensity consistent with PRES.

Sixty hours after caesarean section, the epidural catheter was removed with its tip intact. Following epidural catheter removal, it was noted that clear fluid was draining from the insertion site. Diagnosis of CSF leakage was made based only on glucose testing. At that moment, the patient complained of persistent frontal headache exacerbated in standing position, without any factor of relief, nausea or vomiting. On physical examination BP was normal (128/73 mmHg) and the patient was afebrile. State of consciousness was normal, and focal neurological deficits, including meningeal signs, were absent. It was presumed to be a Post-Dural Puncture Headache (PDPH) secondary to a CSF-cutaneous fistula. After discussion with a neurologist, close monitoring of neurological deficits and treatment with conservative measures were recommended. Therefore, bed rest and supportive therapy such as rehydration, acetaminophen-caffeine, non-steroidal anti-inflammatory (ibuprofen), and antiemetics (metoclopramide and ondansetron) were prescribed and the epidural site was covered with sterile compressive dressing. On continued evaluation (4 hours afterward), the epidural insertion site appeared benign with no further discharge following placement of the dressing, and the patient remained afebrile without neurological symptoms. Despite the instituted measures, 12 hours after the diagnosis of PDPH, the frontal headache persisted in supine position. At this time, BP was high (170/100 mmHg). Approximately 88 hours post-caesarean section the anesthesiology team was called because the patient experienced shortness of breath. There were no other symptoms associated. The patient had desaturation (oxygen saturation of 90% at room air), chest crackles and high BP (150/85 mmHg) on physical examination. Therapy with oxygen and bronchodilators was administered with slight improvement. Ninetytwo hours after caesarean section the patient presented sudden amaurosis followed by two episodes of generalized tonic-clonic seizures. The in-hospital medical emergency team was activated and intravenous diazepam (total of 10 mg) was administered, after which seizures subsided. There were no other apparent focal neurological deficits. At that time, BP remained high (167/100 mmHg) and oxygen saturation was 90% (room air); chest crackles persisted on pulmonary auscultation and blood glucose was normal. After a period of drowsiness due to the postictal period and administration of benzodiazepines, the patient presented psychomotor agitation and had to be sedated with propofol and fentanyl and intubated. After activation, the in-hospital emergency team considered the following differential diagnoses of postpartum headache with visual changes and seizures: drug withdrawal, central nervous system infection, metabolic disturbances (hypoglycemia, hypocalcaemia, hypomagnesemia, hyponatremia or hypernatremia), eclampsia, space-occupying lesions, vasculitis, venous thrombosis, stroke and complications of epidural analgesia (dural puncture, meningoencephalitis, intracranial hemorrhage, cerebral infarction and pneumoencephalus). In order to rule out those diagnoses, laboratorial tests were requested (blood tests, urinalysis and

urine toxicology screening) in addition to CT scan with contrast and MRI arteriography/ venography. Laboratory tests revealed an elevated white cell count of 17,000 and 85% neutrophils, with normal counts for hemoglobin and platelets. C-reactive protein was 5 mg/dl and liver function tests revealed a two-fold increase in aspartate aminotransferase (AST). Renal function tests, electrolyte balance and serum glucose levels were also normal. Urinalysis was remarkable for albuminuria with a urinary albumin-creatinine ratio of 344,3 mg/g. Blood clotting tests were within normal limits. Urine toxicology screening was negative. Emergency brain CT scan revealed diffuse edema and a hypodense lesion in the left parieto-occipital lobe, and MRI Coronal FLAIR image showed a diffuse hyperintense signal in deep white matter and subcortical white matter along the parieto-occipital regions, suggestive of PRES (Figure 1).

The patient had neither a previous/recent history of drug abuse nor positive urine toxicology screening; therefore, drug withdrawal was excluded as a possible etiology. Also, normal laboratory findings ruled out the hypothesis of metabolic disturbances. In this case, the diagnosis of PDPH secondary to CSF-cutaneous fistula was made based only on glucose testing. Presence of glucose in the leaking fluid suggested against a leak of epidural medication. However, this did not allow a differentiation between CSF leakage or interstitial fluid accumulation. In order to distinguish between these fluids, proteins would have to be measured (interstitial fluid generally has a one to two orders of magnitude greater protein concentration compared to CSF). As proteins were not tested, interstitial fluid leakage could not be ruled out in this case.

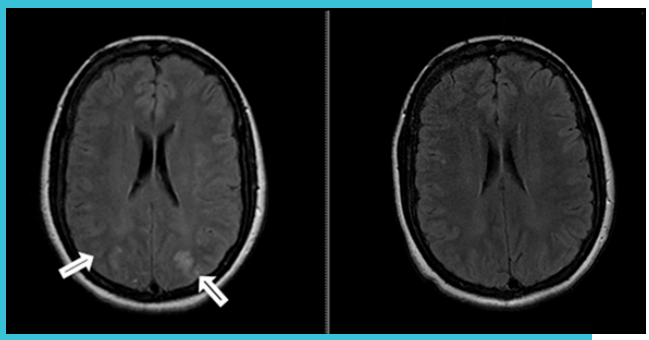
Figure 2.



Time series of the patient's blood pressure during hospital admission. The patient underwent caesarean section on October 12th and the in-hospital medical emergency team was activated on October 16th.

The CT scan and MRI allowed the exclusion of other complications associated with epidural analgesia, as well as space-occupying lesions, vasculitis, venous thrombosis and stroke. Regarding the diagnosis of encephalitis and meningitis, the counter arguments were: absence of signs of meningeal irritation, fever or skin abnormalities on physical examination; and absence of imaging findings suggestive of central nervous system infection. On the other hand, leukocytosis and mild elevation of C-reactive protein supported the hypothesis of encephalitis or meningitis. However, these biochemical markers are nonspecific. Lumbar puncture was not performed because the diagnosis of eclampsia seemed the most likely cause for seizures and visual loss. The diagnosis of eclampsia was based on the clinical course, the detection of a vasogenic edema with no signs of cytotoxic edema in the MRI scan suggestive of PRES, and the combination of increased BP (Figure 2) with proteinuria, acute pulmonary edema and a recent childbirth in the patient's medical history. The obstetric team was involved early and the patient was admitted into the Intensive Care Unit (ICU). The initial treatment comprised intravenous administration of magnesium sulphate (first bolus of 4 g followed by an infusion of 1 g/ 24h) and labetalol. Approximately 6 hours after admission into the ICU, the patient was extubated. Evolution was favorable with the disappearance of headache, seizures and immediate recovery of visual acuity. The patient's BP gradually normalized. Ophthalmological examination revealed fundi suggestive of cortical blindness and no sign of hypertensive retinopathy. A transthoracic echocardiogram was also performed showing normal left ventricular size and systolic function, mild mitral and tricuspid regurgitation, slightly elevated pulmonary artery pressure and no pericardial effusion. One week later, the follow-up MRI (Figure 3) revealed complete resolution of the abnormalities observed in the previous MRI. Ten days after admission into the hospital, the patient was discharged on lisinopril 20 mg daily and without any neurological sequelae. In the ICU follow-up consultation, 3 months after discharge, the patient reported ongoing sleep disturbances, panic attacks and tested positive for anxiety and post-traumatic stress disorder. The patient was referred to psychological treatment. A strategy of brief psychotherapy was adopted. Ophthalmological examination was normal on follow-up, 5 months after discharge.

Figure 3.



T2-Weighted magnetic resonance image obtained before (left) and after (right) treatment of LPE.

Discussion

PRES is a clinical-radiological entity described for the first time in 1996 by Hinchey et al [1]. It is characterized by a variety of presenting symptoms and abnormal CT scan and MRI findings with predominantly posterior cerebral edema, all of which are potentially reversible with appropriate management during the acute phase. Clinical symptoms include headache, nausea, altered mental status, vision loss, paresis, hemianopsia, generalized seizures and coma [2]. Most cases manifest with acute to subacute hypertension. Visual disturbances are the most frequent neurological symptoms encountered [3]. PRES may occur in several situations, although the most common trigger involves an acute elevation of BP [4]. The pathophysiological mechanism entails a failure in the autoregulation of cerebral blood flow with hyperperfusion, passive vasodilation and increased vascular permeability, leading to fluid transudation to cerebral parenchyma and reversible vasogenic edema without infarction. Another proposed mechanism [5] consists in direct injury to the endothelium caused by vasoplegia or immunosuppressive agents, which secondarily leads to break down of the blood-brain barrier and to manifestations of PRES. Classic CT scan findings are those of bilaterally symmetric low attenuation in the posterior parietal and oc-

cipital lobes, whereas MRI demonstrates hyperintensity on T2-weighted images in the same distribution. The most specific evidence for PRES is the demonstration of resolution of vasoconstriction within a 12-week period. Preeclampsia is characterized by hypertension (BP ≥140/90 mmHg on 2 occasions, at least 6 hours apart) and proteinuria during the antepartum and postpartum periods. When associated with a new onset of seizures which is not attributable to other causes, it is termed eclampsia. Usually it occurs between 20 weeks of gestation and 48 hours postpartum. If it occurs after 48 hours and up to 30 days after delivery, it is called late postpartum preeclampsia or eclampsia. LPE accounts for 16% of cases of eclampsia [6] and has been associated with PRES [7]. LPE has a clinical picture different from classic eclampsia in that pregnancy and delivery often are completely normal and without signs of a preeclamptic syndrome, making the diagnosis of LPE more difficult. In 50% to 75% of cases, LPE manifests after a prodrome consisting in headaches of increasing severity days to weeks after delivery. Vision changes appear in 19% to 32% of cases. LPE seizures begin within hours to days of onset. Once the convulsive phase of LPE has begun, T2-weighted MRI often demonstrates findings consistent with PRES [8]. In patients with eclampsia, PRES is more common in the postpartum period owing to an increased accumulation of fluids and tendency to develop cerebral edema. In this patient, the presence of headache with postural features contributed to a diagnostic dilemma. The clinical picture was initially interpreted as PDPH secondary to CSF-cutaneous fistula. Nevertheless, other differential diagnoses should be ruled out in these cases, including LPE. Magnesium sulphate is the preferred anticonvulsant in the management of PRES secondary to eclampsia. Its superiority in the prevention of seizures compared to diazepam and phenytoin was clearly demonstrated by a multicentre trial including 1680 eclamptic patients [9]. Additionally, it is known to have a higher anti-edematous effect than mannitol during PRES. Also, a rapid decrease in serum magnesium seems to contemporarily trigger PRES. Nevertheless, if BP remains high (systolic >160 mmHg or diastolic >110 mmHg) despite the treatment with magnesium sulphate, other drugs such as labetalol, nicardipine, hydralazine or sodium nitroprusside can be used. In the postpartum period, labetalol is considered the preferred intravenous agent, owing to its action on alpha and beta receptors without triggering reflex tachycardia [10]. Whether PRES abnormalities are reversible is still under debate. Delayed recognition and inadequate management of PRES may lead to secondary complications (e.g. status epilepticus, intracranial hemorrhage, ischemic infarction), thus it is of great importance to find the triggering cause and minimize the number of seizures. For this reason, and in order to optimize the diagnostic procedure, close multi-disciplinary cooperation is important. This case report demonstrates that the presence of a headache completely unrelated to or possibly aggravated by a post-dural puncture has to be taken into account as a subtle sign suggestive of mild preeclampsia. Also, it emphasizes the importance of properly and quickly recognizing PRES and LPE, as tardy or inappropriate treatment can lead to irreversible neurological damage.

Acknowledgement
None

Declaration of Interest
None

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