

Unexpected Maternal Convulsion: An Idiopathic Case of Posterior Reversible Encephalopathy Syndrome after Delivery

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ARTICLE INFO	ABSTRACT
<p><i>Article type:</i> Case Report</p>	<p>Posterior reversible encephalopathy syndrome (PRES) is associated with various clinical manifestations such as headache, blurred vision, confusion and tonic-clonic convulsion. Some of the predisposing factors for PRES include hypertensive encephalopathy, preeclampsia and eclampsia, lupus erythematosus, thrombotic thrombocytopenic purpura and long-term use of immunosuppressive drugs. This condition rarely occurs after normotensive and uneventful pregnancies. Several theories have been proposed on the etiology of PRES. For instance, endothelial injury and brain edema have been reported as possible causes of PRES. Although PRES is a temporary condition, proper and timely management of the disorder in the acute phase is critical for the prevention of permanent neurological complications. During pregnancy, PRES is normally accompanied with hypertension. In this paper, we present a rare case of PRES in a normotensive pregnancy in a 25-year-old parturient woman (Gravida 2, Ab 1). The patient unexpectedly manifested symptoms of tonic-clonic convulsion one hour after an uneventful vaginal delivery, which were successfully managed. According to our observations, PRES has various clinical manifestations with unexpected occurrence in some cases. Therefore, it is recommended that maternity centers be well-equipped with resuscitation tools, emergency drugs and expert staff so as to manage unforeseen PRES efficiently and prevent permanent maternal neurological complications and mortality.</p>
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Introduction

Convulsion is a deleterious obstetric emergency that could jeopardize both the mother and neonate. Eclampsia is the most common form of maternal convulsion occurring in 1/2,000 of pregnancies and is generally accompanied with seizure-like presentation, hypertension and proteinuria (1). Incidence rate of eclampsia is comparatively higher during the third trimester of pregnancy; however, it could prevail in the postpartum period. Seizures occurring two days after delivery should be particularly evaluated for other etiologies, such as posterior reversible encephalopathy syndrome (PRES) (2).

PRES was first introduced in a study conducted by Hinchey in 1996 (3), in which one fifth of the patients presented with PRES had

eclampsia associated with other etiologies, including the use of immunosuppressive drugs and hypertensive encephalopathy (3). Among other possible etiologies for PRES are auto-regular disturbances in brain vessels, vasospasm and vasogenic cerebral edema, which normally involve posterior lobes (4). Diagnosis of PRES depends on clinical and radiological findings, and this condition is usually accompanied with other events, such as hypertension (5).

In this paper, we presented a rare case of unexpected PRES associated with none of the common predisposing factors, such as hypertensive encephalopathy, preeclampsia and eclampsia, lupus erythematosus, thrombotic thrombocytopenic purpura and long-term use of immunosuppressive drugs (6).

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Case report

A 25-year-old primiparous woman with term pregnancy (42 weeks of gestation) was admitted to Mobini Hospital in Sabzevar, Iran for delivery. The patient had no medical history of hypertension or epilepsy during and before pregnancy and was scheduled for oxytocin labor induction. Labor induction lasted for 16 hours, and monitoring for vital signs was unremarkable (systolic blood pressure: 100 mmHg, diastolic blood pressure: 60 mmHg).

Other laboratory test results were normal, and labor progression was also acceptable. The patient received pethidine (50 mg) during the active phase of labor (4 hours before delivery), and after a successful vaginal delivery, she gave birth to a female infant with birth weight of 3500 grams. Based on routine labor management, the patient was injected with intramuscular oxytocin (10 mg) after delivery, and placenta was extracted in time. Moreover, uterine bleeding volume was within the normal range.

The patient was alert and could communicate verbally with midwives during episiotomy repair, and her vital signs were normal as well. However, after the termination of delivery process and before transfer to the postpartum room, the patient had a sudden, severe generalized tonic-clonic seizure accompanied with transient apnea and unconsciousness. Necessary procedures, including diazepam injection (10 mg), were immediately performed, and the convulsion was managed successfully. In addition, magnesium sulfate (4 mg) was administered for eclampsia. Vital signs of the patient after these procedures were as follows: blood pressure (BP): 90/60 mmHg, pulse rate (PR): 72, respiratory rate (RR): 14 and temperature: 37°C.

Systemic examination (respiratory, cardiovascular and abdominal evaluation), papillary reactions and fundoscopic examination of the parturient, related laboratory test results were also within the normal range, as follows: Na: 137, K: 3.5, Ca: 9.4, serum glutamic oxaloacetic transaminase (SGOT): 35, serum glutamic pyruvic transaminase (SGPT): 38, platelet: 170,000, urinalysis (UA): normal and proteinuria: 150 mg/24 hour.

Brain computed tomography (CT) was

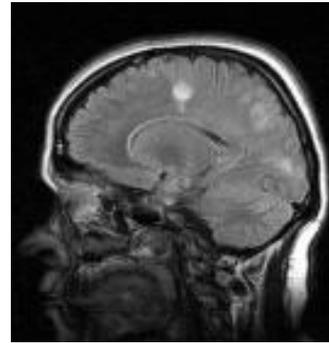


Figure 1. Abnormal Signal Intensity in Magnetic Resonance Imaging (Subcortical Area)

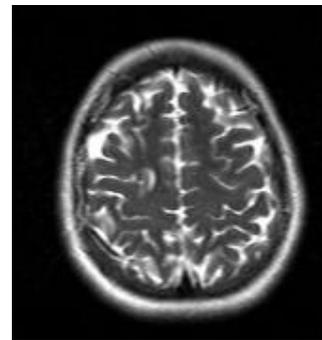


Figure 2. Abnormal Signal Intensity in Magnetic Resonance Imaging (Paraventricular Area)

normal, and no consolidation was present. Moreover, magnetic resonance imaging (MRI) revealed abnormal cortical and subcortical signal intensity involving the parieto-occipital lobes and paraventricular regions in T2-weighted and fluid attenuation inversion recovery (FLAIR) sequences (Figure 1 and 2), which confirmed the diagnosis of PRES. During seizure management at the hospital, the patient responded to phenytoin injection and had no other seizure-associated manifestations.

The patient was discharged three days after delivery in good overall condition, and phenytoin was prescribed as outpatient medication. After a 24-month follow-up, the patient reported no episodes of seizure, and resolution of PRES was confirmed. It is noteworthy that informed consent was obtained from the patient to report her case.

Discussion

PRES is a reversible condition associated with various clinical manifestations. Common symptoms of PRES include headache, nausea and vomiting, blurred vision, blindness and tonic-

clonic convulsion, which could be accompanied by hypertensive status during pregnancy. PRES may rarely occur after normotensive and uneventful pregnancies, which is described as atypical in the literature (7).

Etiology of PRES is under debate; the most acceptable theory declares that rapidly developing hypertension leads to a breakdown in cerebral autoregulation, especially in posterior lobes, leading to PRES. Following this event, fluid and protein extravasation could give rise to vasogenic cerebral edema. Furthermore, an alternative theory considers endothelial damage as a possible cause of preeclampsia, eclampsia and sepsis. Another theory suggests vasospasm with subsequent brain ischemia as a major predisposing factor for PRES (4).

Some studies conducted in this regard have reported that pregnancy alone may affect brain vessels and lead to PRES. Moreover, animal studies during late pregnancy have indicated vasoconstriction reactivity in response to serotonin, which was inconsistent with the results reported in non-pregnant cases; however, this is in line with the theory of vasospasm regarding the incidence of PRES (8).

Differential diagnoses of PRES during the peripartum period are eclampsia, subarachnoid and intracranial hemorrhage, brain thrombosis, epilepsy, meningoencephalitis and amniotic fluid embolism (6, 7). Eclampsia occurs during the postpartum period in 5-26% of women; therefore, differentiating this condition from PRES is of paramount importance since the symptoms are not specific, and there is the possibility of misdiagnosis (9).

Accurate diagnosis of PRES largely depends on imaging procedures. CT-scan is usually normal, while MRI is considered as the main diagnostic measure. Specific findings related to vasogenic cerebral edema could be distinguished in MRI-FLAIR images. In addition, detection of hyperintensive signals, which are more apparent in subcortical white matter of the posterior lobes, would be a key element in the diagnosis of PRES (4, 6, 10). In one study, McKinney et al. discussed the incidence of PRES, and reported the endemic regions as parieto-occipital (98.7%), temporal (68.4%), thalamus (30%), cerebellum (36.4%) and brain stem (18.4%) areas (11). These findings are

consistent with the results obtained by the present study, which indicated the parieto-occipital area as the most significant region affected by PRES in MRI.

Multidisciplinary approach is normally used for the treatment of PRES. Management of seizures via different methods, such as anticonvulsant therapy with phenytoin, magnesium sulfate or antihypertensive drugs, should be taken into account (4, 7, 10, 11). In the present study, antihypertensive therapy was not performed, while Cho et al. used labetalol and magnesium sulfate infusion for the treatment of PRES, which resulted in the successful stabilization of their patient (10). In another study, Legiel et al. estimated that 35-40% of PRES patients may require intensive care unit admission and mechanical ventilation (5).

In their research, Barrent et al. reported that 35-100% of the cases presenting with PRES return to baseline condition, while 7-8% may expire (12). Several studies in this regard have emphasized that treatment of PRES with appropriate methods during the acute phase of the condition plays a pivotal role in the prevention of adverse maternal outcomes and achieving full recovery (7, 11, 12). Hypertension is considered as a significant cardiovascular risk factor in both men and woman. In their study, Shakutala et al. described a case of normotensive pregnancy followed by the incidence of PRES accompanied with hypertension (220/120 mmHg) on day four of postpartum (7, 13). Surprisingly, the patient in our study remained normotensive through all the phases of seizure management.

In conclusion, PRES is a life-threatening, yet reversible, condition. In the presence of prodromal symptoms of eclampsia, peripartum convulsions are likely to occur. Therefore, consideration of PRES as a rare and sudden phenomenon could be effective in the prevention of maternal and fetal complications. Furthermore, appropriate management of PRES in the acute phase could result in complete recovery of patients. On the other hand, inaccurate approaches may lead to permanent neurological complications and even death. Owing to the fact that PRES has various clinical manifestations and could sometimes occur unexpectedly, it is recommended that maternity

centers be well-equipped with resuscitation tools, emergency drugs and expert staff so as to manage unexpected PRES.

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Conflicts of Interest

The authors declare no conflicts of interest.

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