Status epilepticus caused by cerebral venous thrombosis in the puerperal period: case report and literature review

Orivaldo Alves Barbosa, João Filho Araujo Ribeiro, Paola Lima Lemos

Abstract

Status epilepticus (SE) caused by cerebral venous thrombosis (CVT) is rare in pregnancy and puerperium, with few cases described in literature. Seizures in pregnancy are usually due to previous epilepsy or pregnancy related disease, such as eclampsia, posterior reversible encephalopathy syndrome (PRES), reversible cerebral vasoconstriction syndrome (RCVS), eclampsia and thrombotic thrombocytopenic purpura (TTP). Due to its high mortality, the SE and the underlying disease must be promptly managed. In this article, we present a case of SE in postpartum due to cerebral venous thrombosis and we reviewed the cases described on the topic.

Key words: Status epilepticus, puerperium, cerebral venous thrombosis.

Introduction

Status epilepticus (SE), defined as 5 minutes or more of continuous seizure or two or more seizures without recovery of consciousness, is a life-threatening neurologic disorder. In pregnancy and postpartum, SE is exceedingly rare, with a high maternal and fetal mortality rate. (1)

In this article, we present a case of SE in postpartum due to cerebral venous thrombosis (CVT) and we reviewed the cases described on the topic. CVT should always be a consideration in pregnant and postpartum women with focal neurological signs, including SE.

Case report

This is a case report of a patient of the Intensive Care Unit of the General Hospital Dr. César Cals de Oliveira in the year 2016, and the patient is currently accompanied as outpatient. Patient authorized the publication of his case, knowing that she is free of any costs and that her identity will be preserved. The data were collected from the medical record and the interview with the patient. The project was submitted and approved by the Ethics Committee in Research of the Federal University of Ceará, following the norms of Resolution 510/2016, of the Brazilian National Health Council.

A 24-year-old female patient from the Brazilian Northeast presented with elevation of pressure levels, drowsiness and visual turbidity at the end of pregnancy. She sought care in the thirty-eighth week of pregnancy being diagnosed with hypertensive disease and indicated termination by cesarean delivery with improvement of symptoms. On postpartum day ten she had generalized seizures and was rushed to our hospital. She arrived 60 min later and had lost consciousness, and she still presented with a generalized tonic-clonic seizure (GTCS). A dose of intravenous diazepam and magnesium sulfate were administered, without resolution, followed by phenytoin, wich resulted in transient seizure control over the next 12 h. The patient was transferred to the Intensive Care Unit. In the next 24 hours 2 new seizures occurred and a loading dose of valproate was administered with control of the symptoms.

A cranial computerized tomography (CT) showed a hyperdensity in superior sagittal sinus (dense clot sign) and two foci of hemorrhage were noted in the

From Internal Medicine Department, Hospital Cesar Cals, Centro, Fortaleza, Ceará, Brazil (Orivaldo Alves Barbosa), Neurology Department, Hospital Cesar Cals, Centro, Fortaleza, Ceará, Brazil (Paola Lima Lemos), and Hospital Walter Cantidio, Fortaleza, Ceará, Brazil (João Filho Araujo Ribeiro).

Address for correspondence:
Orivaldo Alves Barbosa
Internal Medicine Department, Hospital Cesar Cals
Avenida do Imperador, 545, Centro, Fortaleza, Ceará, Brazil
Mobile phone: 5585 81817009
Fax: 5585 31015333
Email: orivaldo.alves.barbosa@gmail.com

frontoparietal area (**Figure 1**). An urgent brain magnetic resonance imaging (MRI) and venogram (VRM) demonstrated a lack of flow in superior sagital sinus (**Figure 2**). The patient was diagnosed with cerebral venous thrombosis and hemorrhagic infarctions. An investigation of thrombophilias, including nocturnal paroxysmal hemoglobinuria, mutation of the prothrombin gene and Leiden's factor V, serum homocysteine, antiphospholipid antibody syndrome, and protein C and S deficiency, were performed, with normal results.

Anticoagulation was initiated with enoxaparin 120 milligrams per day and warfarin 5 milligrams, maintaining international normalized ratio (INR) between 2.5-3.5. After seven days the patient was discharged asymptomatic with valproate 1.5 grams per day and warfarin, remaining asymptomatic one year after the event.

Discussion

Seizures and SE in pregnancy can be divided into three categories: (2) 1) patients with established disease, such as epilepsy, decompensating in pregnancy and puerperium, usually by decreasing the serum level of antiepileptic drugs (AED) due to hypervolemia of pregnancy and induction of hepatic metabolism; (1) 2) new onset non-pregnancy related seizure disorders, such as brain tumors, hypoglycemia; 3) pregnancy related seizures, caused by CVT, posterior reversible encephalopathy syndrome (PRES), reversible cerebral vasoconstriction syndrome (RCVS), eclampsia and thrombotic thrombocytopenic purpura (TTP). In this context, SE is rare, which has been reported to occur in 0.6% of all patients with epilepsy during pregnancy. (3) A retrospective study in a Neurological ICU identified seven cases of SE in pregnancy and postpartum, with a mortality rate of 28.5%. (1) The causes included CVT, limbic encephalitis, and AED withdraw. Other case reports describe brain tumors, RCVS, as systemic lupus, vitamin B6 deficiency, cavernous angiomas and herpes encephalitis as potential causes. (4-8)

There are no specific guidelines for this condition in pregnancy; the pharmacological management is extrapolated from studies in non-pregnant patients, usually by controlling the seizures with AEDs followed by anesthesia in refractory cases.

Although rare as a cause of stroke overall, CVT has high incidence during pregnancy and postpartum, especially during the first trimester in women with an underlying thrombophilia and in puerperium. Most patients with CVT present with headache; however, in 40% of patients seizures can occur. (9) The frequency of peripartum CVT is 11.6 cases per 100,000 deliveries in countries, although exact data for Latin America countries is uncertain. (10)

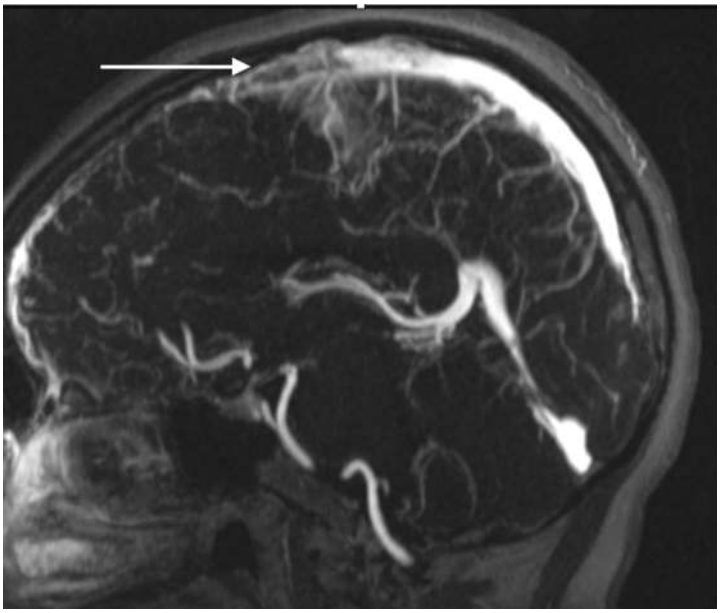
Even with hemorrhage, CVT is treated by anticoagulation. During pregnancy, therapeutic doses of low molecular weight heparin. Data from non-pregnant patients suggest that 80% of patients have full recovery and that the recurrence rate is under 10%. (10)

Conclusion

SE, although a rare event in pregnancy and puerperium, must be promptly managed due to its high mortality rates. The obstetrician and intensive care physician must be aware of the risk factors, causes and complications of this condition. CVT, although uncommon, must always be considered in any patient with focal neurological signs in pregnancy and puerperium.

Figure 1. Cranial computed tomography without contrast administration showed a hyper-density in superior sagittal sinus (dense clot sign [arrow]) and two foci of venous haemorrhage were noted in the frontoparietal area

Figure 2. Magnetic resonance 2D time of flight (TOF) venography obtained without the administration of contrast material revealed the absence of a signal in the superior sagittal sinus (arrow), compatible with a filling defect caused by thrombus



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