Medicine & Clinical Science

Case Report

Neuropsychiatric Manifestations, As a Rare Presentation of Venous Sinus Thrombosis Two Weeks After Cesarean Section: First Report in the Gambia

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Abstract

Cerebral venous sinus thrombosis (CVST) is a rare and potentially fatal condition, accounting for about 0.5% of all cerebrovascular disease cases globally. It is challenging to diagnose due to its rarity and diverse clinical presentations that can mimic other acute neurological conditions [1]. The reported worldwide incidence of CVST during pregnancy and postpartum is approximately 3 to 4 cases per million, with superior sagittal sinus thrombosis (SSST) being the most prevalent type. Risk factors include the prothrombotic state of pregnancy in the third trimester, obesity, accidental dural puncture, deficiencies in protein C and S, antiphospholipid syndrome, and the use of oral contraceptives among others. Symptoms typically manifest within the first three weeks postpartum, with severe headache being the most common complaint. The low number of reported cases in sub-Saharan Africa suggests underdiagnosing or underreporting, with infection being the primary cause in this region. We report a case at our facility who, two weeks after a cesarean section, presented with severe headache, confusion, irritability, restlessness, and later developed a focal motor deficit. Imaging studies were crucial in diagnosing SSST and ruling out other conditions causing acute brain injury. This is the first reported case in The Gambia, and one of the few described in West Africa related to an obstetric procedure. The onset, characterized by neuropsychiatric symptoms, is unusual and interesting, as these manifestations are less common than the typical headache, seizure, and focal motor deficit described in the literature.

Introduction

30-year-old female patient with a history of previous cesarean section arrived at the emergency department with a severe headache, intense restlessness, and moderate disorientation. Normal Initial medical examination, except for irritability and restlessness. Admitted at the ICU, under sedation, with the preliminary diagnosis of puerperal psychosis. Over the next 12 hours, she developed left-sided hemiparesis with muscle strength 4/5 (upper and lower), transient bilateral vision loss, with GCS of 12/15. Vital signs remained stable. Treatment began with anti-cerebral edema and neuroprotection measures, along with a request for a brain CT scan. The axial tomography revealed thrombosis of the superior sagittal sinus [Fig1-3]. Subcutaneous enoxaparin initiated, leading to gradual but significant progress. Further laboratory studies ruled out autoimmune diseases, infections, antiphospholipid syndrome, or other hypercoagulable conditions. As a result of her neurological improvement, the medical team decided to transfer to an open ward and discharged on the seventh day, without any central nervous system complications. With a GCS of 15/15 and NIHS/0. The final diagnosis upon discharge was sagittal sinus thrombosis related to the previous cesarean section surgery.

Discussion

Cerebral venous sinus thrombosis is a rare but life-threatening disorder, accounting for 0.5% to 1% of all stroke cases globally. It is particularly prevalent in fertile women, being three times more common than in men, often linked to factors like contraceptive pill use, pregnancy, and the postpartum period. The condition is associated with the deterioration of cerebral venous sinuses during childbirth, influenced by intracranial pressure instability and hypercoagulability in pregnancy. Diagnosing it can be challenging due to its similarity to other cerebrovascular diseases and conditions related to pregnancy, such as postpartum preeclampsia, post-spinal puncture headache,

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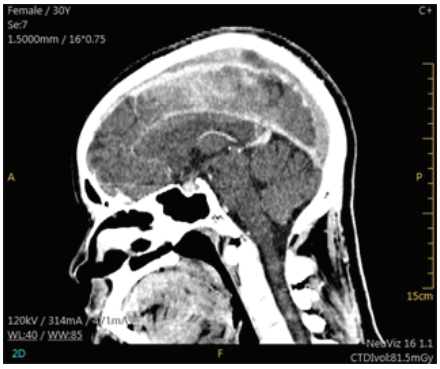

Figure 1. *Sagittal view*

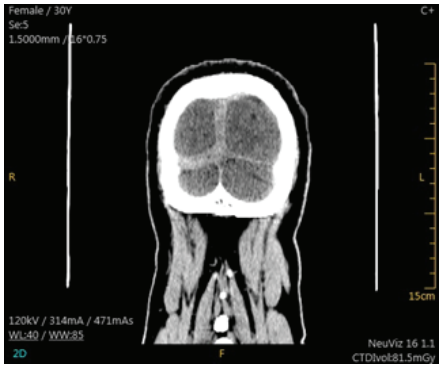

Figure 2. *Coronal view*

and CNS vasculitis, including neuro Behçet's. Risk factors include estrogen therapies, thrombophilia, hypercoagulability, infections, autoimmune and autoinflammatory CNS conditions. Common abnormal laboratory findings include elevated fibrinogen levels and various serum lipoproteins. Neuroimaging studies show that the superior sagittal sinus and transverse sinus are frequently affected locations [2-4].

Patients with CVST often present with headaches, seizures, and signs of focal neurological deficits. Clinical symptoms progress from the blockage of the venous sinuses, cortical veins, jugular veins, or internal cerebral veins of the cerebral venous sinuses and compromised drainage of cerebrospinal fluid. Thrombosis of the cerebral veins results in increased venous and capillary pressure, decreasing cerebral perfusion, as well as impaired cerebrospinal fluid absorption, ultimately causing increased intracranial pressure [5,6]. Clinical symptoms typically emerge within the first three weeks postpartum in most cases, with severe headache being the most common complaint (82.1%). However, the International Classification of Headache Disorders 3rd edition (ICHD-3) notes that SCVST headache lacks specific characteristics. Nevertheless, it is usually accompanied by nausea, seizures, altered level of consciousness, papilledema, and focal neurological deficits. Reports indicate that 25% of patients experienced blurred vision, 33% were unconscious upon admission, 13% had hemiplegia, and 33% had fever [7]. Cranial nerve syndromes associated with CVST include vestibular neuropathy, unilateral deafness, facial weakness, diplopia, and some patients may exhibit isolated neuropsychiatric symptoms, such as delirium, psychosis, depression, anxiety, irritability, personality change, apathy/abulia, or cognitive decline, which can be particularly misleading like our case [8].

Differential diagnosis

In our patient, the association of headache, neuropsychiatric symptoms, bilateral but transient vision loss with focal neurological manifestations complicated the establishment of the differential diagnosis. The presence of numerous potential diagnoses due to clinical similarities with other causes, the likelihood of overlapping symptoms, or coexistence with other conditions made complex the approach to the patient. Demanding to rule out other cerebro vascular diseases initially, ischemic (thrombotic or embolic) either or hemorrhagic. Others condition could be considered such as: Posterior reversible encephalopathy; Postpartum eclampsia, as well as less common like Neuro Behçet's or other forms of vasculitis [9]. Postpartum-related CVST typically has a more sudden onset and a better prognosis compared to CVST from other causes. CVST, often manifesting as superior sagittal sinus thrombosis, is more prevalent in the postpartum period than during pregnancy. New predisposing factors such as obesity, COVID-19, and COVID -19 vaccine-induced thrombocytopenia have been identified [10-12].

Postpartum preeclampsia is a rare but severe condition linked to high blood pressure after childbirth. It typically occurs within 48 hours but can also develop up to six weeks post-delivery, potentially leading to strokes, seizures, and other complications if not promptly addressed. Posterior reversible encephalopathy syndrome (PRES)is a clinical-radiological disorder characterized by encephalopathy, seizures, headaches, and visual disturbances. It is associated with conditions that cause damage to the endothelium. The incidence is currently unknown, and it has traditionally been linked to eclampsia, hypertensive encephalopathy, immunosuppressive therapies, renal failure, connective tissue disorders, or sepsis [13-16]. The hallmark radiological feature of vasogenic cerebral edema was not observed in our neuroimaging study, additionally the absence of clinical conditions associated to PRES, like high blood pressure and impairment of the kidney function, excluded the diagnosis. Digital subtraction angiography (DSA) has been considered relevant for the diagnosing, but its use has declined due to its invasive nature and lack of experienced angiographic skills, contrast-enhanced MRV, gradient-recalled echo, or susceptibility-weighted imaging sequences are the recommended techniques for the diagnosis of cortical venous thrombosis. [17].

Epidemiology

Epidemiology has experienced significant changes in the past decade. The disease's incidence has risen in developed countries from 0.2-0.5 cases/100,000 inhabitants/year to 1.32-1.75/100,000/year, leading to the identification of rare and severe cases [18,19]. Investigations into maternal deaths in the UK have pinpointed thrombosis and thromboembolism as the primary cause of death in women who pass away during pregnancy or within six weeks of its conclusion [20]. The incidence in populations in developing countries remains unknown, primarily due to the rarity and variety of CVST manifestations linked to pregnancy and the absence of neuroimaging studies. However, recently, the diagnosis has increased in several developed countries, mainly due to enhanced understanding of the disease and the availability of imaging diagnostic tools. Multiple case series have been documented in India, China, Senegal, and South America, with the highest incidence reported in the first. Nevertheless, etiology maintains similarities with the pattern observed in developed countries, been the sagittal

sinus continues also the most affected. Conversely, reports are still limited in other developing countries, particularly in sub-Saharan Africa, where data on incidence, prevalence, and epidemiology remain scarce and significantly differ from those reported elsewhere, establishing infectious etiology as the most common origin [21-24].

Treatment

Timely diagnosis and treatment are crucial for achieving better outcomes in pregnant women with CVST. The goals of anticoagulation therapy in CVST are to prevent thrombus growth, aid in re-canalization, and prevent recurrency. General management includes anti-cerebral edema and neuroprotection, then anticoagulant therapy and endovascular thrombolysis have shown effectiveness in treating CVST. The standard initial approach for treating CVST involves intravenous heparin followed by a switch to oral VKAs for 3 to 12 months, depending on the underlying cause, or indefinitely in cases of thrombophilia or recurrent CVST. Based on open-label retrospective and prospective randomized studies, DOACs seem to be a safe and effective alternative to VKAs [17].

Conclusion

Cerebral venous sinus thrombosis (CVST) is a rare and potentially fatal neurological emergency that occurs more often in women during pregnancy and the postpartum period than in the general population. Early diagnosis is crucial due to the potential for full recovery. Considering the absence of specific symptoms, maintaining a high level of clinical suspicion is important. CVST should be considered as a differential diagnosis and treated as an emergency in any woman presenting with typical or vague neurological symptoms, or even just a severe headache, during the postpartum period. In sub-Saharan Africa, the lack of diagnostic resources and the similarity of clinical presentations to other conditions pose challenges and can lead to under-recognition and underreporting of cases. For pregnant women with CVST, full anticoagulant doses of LMWH should be continued throughout pregnancy, followed by LMWH or VKA with a target INR of 2 to 3 for at least 6 weeks postpartum.

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