CASE REPORT

Persistent headache in a postpartum patient: the investigation and management

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SUMMARY

Postdural puncture headache (PDPH) is the most common complication of obstetric regional anaesthesia and the most likely cause of headache in a woman who underwent epidural anaesthesia during delivery. Cerebral venous sinus thrombosis (CVST) is an uncommon cause of postpartum headache. Anaesthesia in obstetrics may lead to long-lasting intracranial hypotension resulting in CVST. CVST is a serious pathology with high mortality if misdiagnosed, but its correct and rapid diagnosis offers the opportunity for early treatment. Cranial magnetic resonance imaging (MRI) is an important modality in the diagnosis of both CVST and intracranial hypotension. The latter condition may be treated either by an epidural blood patch or bed rest and hydration. We report a case of a 36-year-old woman who developed CVST and multiple venous infarcts after an attempted epidural procedure during delivery. She was treated conservatively with bed rest, hydration and low-molecular-weight heparin and the patient recovered completely.

BACKGROUND

Delivery with spinal and/or epidural anaesthesia has become popular recently. Although it is considered safe and effective, epidural analgesia may cause a variety of neurological complications, the most common ones being, accidental dural puncture and postdural puncture headache (PDPH).[1] Headache after inadvertent penetration of the dura with an attempted epidural puncture for anaesthesia is a known common complication but ensuing cerebral venous sinus thrombosis (CVST) is rare. CVST may result from intracranial hypotension, which may be either spontaneous or secondary to spinal procedures such as lumbar puncture (LP) and spinal interventions. The incidence of CVST during pregnancy has been reported in the range of 1:10.000–1:25.000.[2 3] It is a common complication due to dural puncture and cerebrospinal fluid (CSF) leakage.

The triggering factor for CVST is the venous congestion due to a CSF leak through the dural tear. The loss of CSF causes a compensatory venous dilation and a decrease in blood flow. Thus there may be an increased tendency for thrombus formation.[4]

CVST may present with a broad spectrum of different neurological symptoms, including headache. Especially in cases with atypical clinical presentation and in which PDPH is thought to be responsible for current headache symptoms after regional anaesthesia, CVST can easily be diagnosed wrongly or late.[1] Early diagnosis of PDPH leads to adequate and timely therapeutic approaches like an epidural blood patch or bed rest and oral/intravenous hydration. Magnetic resonance imaging (MRI) is accepted as a primary diagnostic method with its high resolution and ability to localise lesions accurately. Its combination with MR venography (MRV) is particularly sensitive for CVST.[5]

In this paper, we present a case of a young woman who had undergone an attempted epidural analgesia for labour and developed PDPH. The headache persisted along with other neurological symptoms. The MRI and MRV findings were consistent with CVST and multiple venous infarcts. The patient recovered completely with a conservative approach. We discuss the diagnosis and management of this patient and review the literature.

CASE PRESENTATION

A 36-year-old woman presented to the neurology outpatient clinic with headache and recurrent seizures. Ten days before presentation, she had undergone a caesarean section. Initially, the procedure had been planned to be performed with epidural anaesthesia; but the epidural catheter placement was problematic and as epidural access could not be achieved due to multiple unsuccessful painful attempts, the procedure had been performed under general anaesthesia. On the first postpartum day, she had suffered from headache and neck spasms. The headache had a frontooccipital distribution. She had been afebrile and her arterial blood pressure had been normal. There was no evidence of focal disorder on neurological examination. She responded well to bed rest, parenteral myorelaxant (intramuscular thiocolchicoside 4 mg/2 mL, twice a day) and oral hydration treatment for a suspected PDPH. On the fourth postpartum day, the headache had relapsed and worsened progressively. Moreover, she had four simple partial tonic seizures with secondary generalisation starting from the right side of her body. This condition had aroused suspicion that a more serious intracranial pathology might be present and a diagnostic MRI had been planned. She had been prescribed carbamazepine 400 mg/day. As an intracranial tumour had been suspected on MRI examination which had been performed at another institution, she was referred to our emergency department. Her physical and neurological examination, optic nerve head appearance, arterial blood pressure and biochemical and coagulation tests were normal. She was without fever. An cranial MRI was performed urgently. Lesions involving the subcortical area and lateral cortex of the right temporal lobe and the

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paracentral area of the left parietal lobe at the convexity were present in the non-enhanced cranial MRI (figure 1A,B). The lesions were hypointense and hyperintense on the T1-weighted images and T2-weighted images, respectively. Vasogenic oedema was present around the lesions. Contrast-enhanced images revealed dural thickening (figure 1D,E). Diffusion-weighted imaging detected acute ischaemia. In addition, heterogeneous intensities were present in the superior sagittal sinus on the sagittal T2-weighted images (figure 1C). A follow-up MRV confirmed thrombosis of the superior sagittal sinus (figure 1F). These findings were found to be consistent with venous sinus thrombosis secondary to intracranial hypotension and venous infarct.

TREATMENT

She was treated conservatively with intravenous fluid treatment and bed rest. Carbamazepine 400 mg/day and enoxaparine sodium 0.6 g/day were administered. An epidural blood patch was not performed. After 2 weeks, she recovered completely and a control MRI showed partial regression of the infarct areas (figure 2A–C) and complete resolution of the dural thickenings (figure 2D–F). She was prescribed carbamazepine 400 mg (q12 h) and acetylsalicylic acid 300 mg (q24 h) and was discharged from hospital.

OUTCOME AND FOLLOW-UP

Three weeks later, her neurological examination was normal. One month after discharge, MRI showed right temporal and left parietal infarcts resulting in encephalomalacia (figure 3).

DISCUSSION

Headache is common during pregnancy, with the primary headaches being 20 times more frequently encountered than the secondary ones.[1 6] Primary headaches are migraines or tension-type headaches. Secondary causes of headaches are intracranial pathologies such as CVST, stroke, intracranial haematoma, meningitis or cerebral tumours.[1]

Pregnant and postpartum women after vaginal delivery especially carry a high risk of PDPH. This condition is more common in women between the ages of 18–30 and with a low body mass index, pre-LP headache and a history of PDPH. But there is no known association between migraine headaches and the increased incidence of PDPH.[7] Migraine tends to improve during pregnancy but can recur early after delivery. Headache is throbbing and often unilateral. It can be accompanied by nausea, vomiting and photophobia, but can rarely present with focal neurological symptoms.[8]

In experienced hands, the incidence of dural puncture is 0.16–1.3%, while the incidence of PDPH is 16–86%.[9] This is a common case in neurological practice which may result in postural headache without fever or a stiff neck and without focal signs. It is usually treated with bed rest and fluids, and an epidural blood patch in severe cases. PDPH is a pain localised in the frontal, frontotemporal or occipital region and typically deteriorating with walking and improving in the supine position. It occurs within 48 h after dural puncture. Associated symptoms are nausea, vomiting and neck stiffness. In addition, some ocular (photophobia and diplopia) and auditory (tinnitus, hyperacusis) symptoms may occur.[9] When the headache is not postural and is progressively worsening, aggravated by coughing, sneezing, straining and awakening the patient during the night, as in this case, further investigation is required. These are the distinguishing features of increased intracranial pressure versus a low-pressure headache.

In recent years, dural puncture during labour has also been reported as an important risk factor for CVST.[10–12] Dural puncture causes a loss of CSF and a decrease in CSF pressure. Loss of CSF results in compensatory vasodilation, which in turn leads to an increase in intracerebral blood volume and a decrease in blood flow. Stasis of blood flow occurs and eventually may lead to thrombosis.[4 12 13] The first case with epidural anaesthesia-associated dural sinus thrombosis was reported in 1986.[14] There are few CVST cases in the peripartum period with prior dural procedure in the literature.[4 8 11–13 15] The pathophysiology of CVST in the postpartum period is not clear. New neurological diseases can be precipitated by pregnancy as a result of the physiological alterations accompanying pregnancy. Venous sinus injury due to the fluctuations in the intracranial pressure during pregnancy, a relative hypercoagulable state due to the increased levels of coagulation factors and activity of thrombocyte adhesion contribute to increased CVST risk during the peripartum period.[12] The other risk factors for CVST are genetic and acquired prothrombotic diseases, head trauma and haematological processes such as leukaemia and thrombocytopenia.[10–12] In our patient, systemic diseases and specific coagulation disorders were excluded by using immunological and specific laboratory tests.

Canhao *et al*[16] reported a significant correlation between the level of decrease in the intracranial pressure induced by LP and the level of decrease in the mean blood flow rate in the sagittal sinus. Another study showed that an immediate decrease in the CSF pressure after dural puncture activated the adenosine receptors in the brain resulting in venous and arterial dilation, responsible from PDPH.[17 18]

Headache as a single symptom of CVST or serious intracranial events in the postpartum period can mimic PDPH. Many CVST patients in the postpartum period were first presumed to have headache secondary to dural puncture and treated with an epidural blood patch. Therefore, potentially fatal cranial peripartum complications can easily be delayed or misdiagnosed, especially in parturients with regional analgesia.[1 19] On the other hand, if the headache persists, changes in character recur despite treatment, or when additional neurological findings develop; brain imaging is necessary and venous thrombosis may be diagnosed.[12]

CVST in women increases the risk of stroke and cerebral haemorrhage may occur if not treated immediately. Haemorrhagic infarcts are independent predictors of adverse outcomes; early diagnosis and treatment are extremely important. Late or incorrect diagnoses are common in CVST due to low incidence, insidious onset and various clinical presentations. Although the most common symptom is headache, it is usually ignored during pregnancy or in the postpartum period. However, rapid advances in imaging technology allow an effective approach for early diagnosis.[5] Cranial computed tomography (CT) without contrast enhancement may have nonspecific findings or may be even normal in 20% of cases. Cranial MRI, being more sensitive, may diagnose about 90% of the patients; but the application of both cranial MRI and MRV together provide a correct and reliable diagnosis in nearly all patients with thrombosis or infarct.[11]

Many of the postpartum CVST cases in the literature have been confined to single localised infarctions.[4 8 12 13] One study reported CVST without infarct.[11] Our case was different from others in the literature in that it had multiple infarcts.

Loss of CSF causes compensatory vasodilation and increase in intracerebral blood volume and the primary objective of treatment of CVST is to decrease the intracranial pressure and to avoid extension of the thrombus.[4 13] Most of the cases reported in the literature were treated with an epidural blood

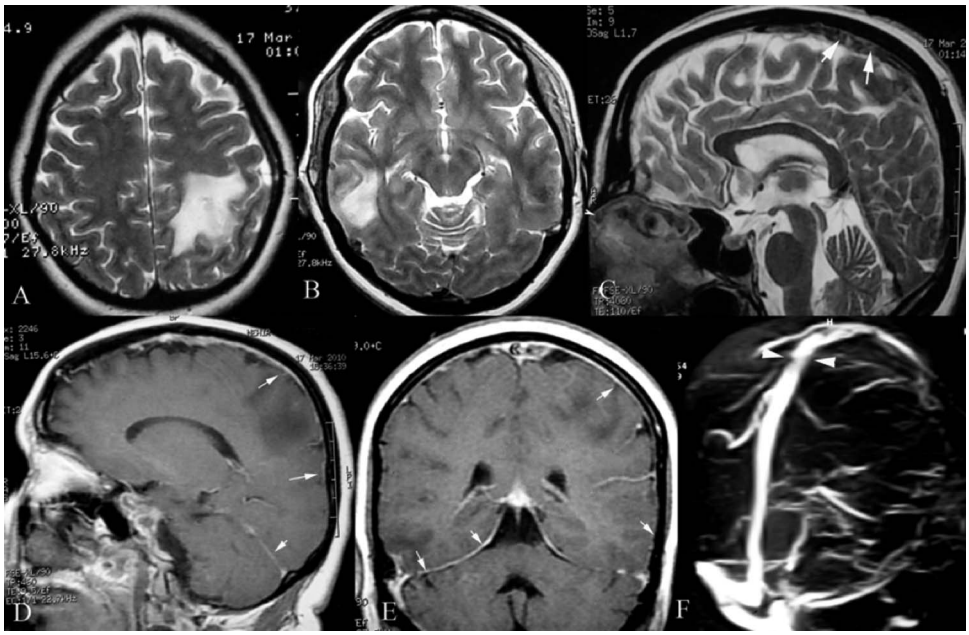

Figure 1 Axial T2-weighted images showing infarcts in the paracentral area of the left parietal lobe at the convexity (A) and in the subcortical area and lateral cortex of the right temporal lobe (B). Sagittal T2-weighted MRI showing a thrombus in the superior sagittal sinus (C, arrows). Sagittal and coronal contrast-enhanced T1-weighted images showing dural thickening and contrast enhancement secondary to the intracranial hypotension syndrome (D and E, arrows). MRV showing thrombosis of the superior sagittal sinus (F, arrowheads).

patch.[4 8 11–13] Ghatge *et al*[13] suggested that an epidural blood patch is frequently required since conservative treatment was inadequate alone. According to some authors, recurrence after an epidural blood patch may occur at all.[8] Some obstetrical studies have demonstrated lower success rates for epidural blood patch as the dural hole made by an 18-gauge needles results in great leakage of CSF.[20 21] A potentially high rate of spontaneous intracranial bleeding makes the safety of anticoagulation therapy a matter of concern among clinicians. However, the European Federation of Neurological Societies (EFNS) has indicated that anticoagulation treatment can be effective in the prevention and treatment of extensive CVST and should therefore be administered.[22] Our patient made a complete recovery in terms of both symptoms and radiological findings after bed rest and oral/intravenous hydration combined with anticoagulation without epidural blood patch. Zupan *et al*[1] also showed that, in those cases with cerebral venous thrombosis and subdural haematomas in the postpartum period following epidural or spinal analgesia

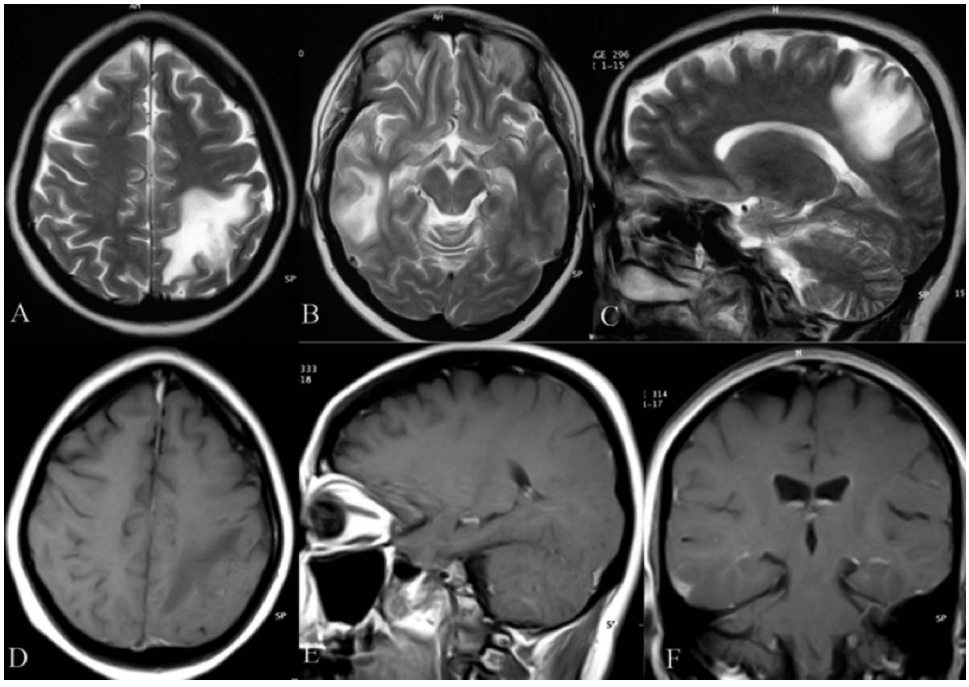

Figure 2 MRI in the postpartum 15th day: axial and sagittal T2-weighted images showing infarcts (A–C). Axial, sagittal and coronal contrast enhanced T1-weighted images showing complete resolution of the dural thickening (D–F).

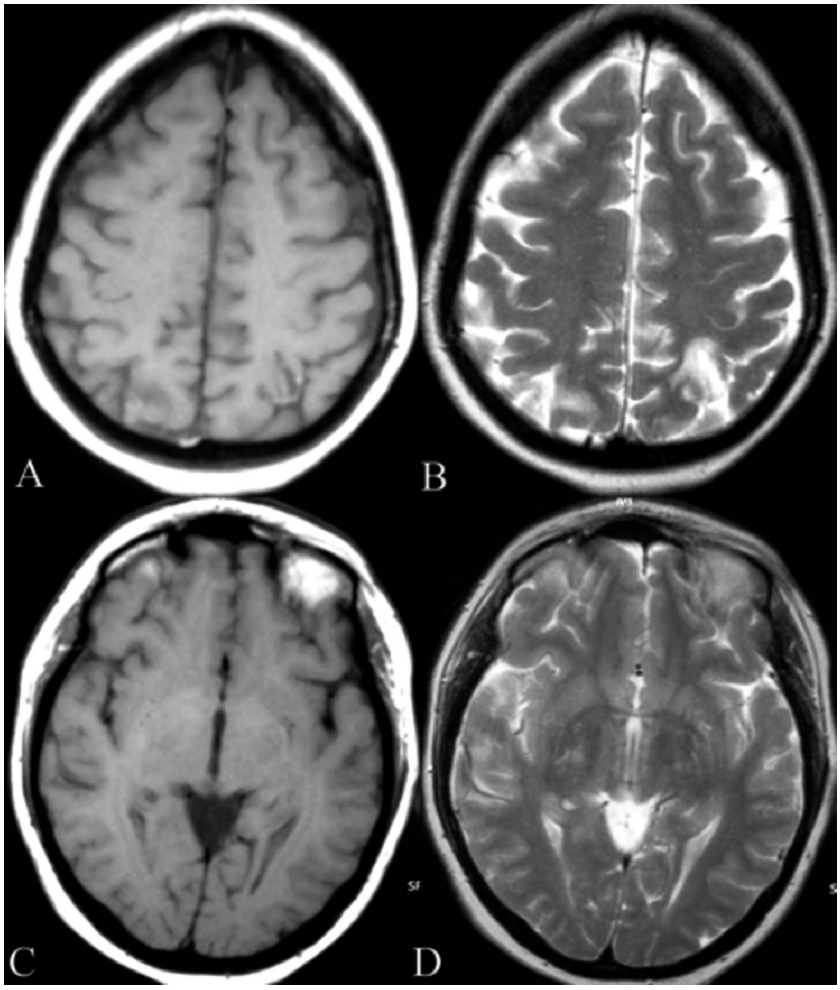

Figure 3 Follow-up axial T1-weighted and T2-weighted images at the first month showing encephalomalacia areas in the left parietal lobe (A and B) and in the right temporal lobe (C and D).

and anaesthesia between 1989 and 2012, most of the patients recovered with conservative treatment.

Early diagnosis of postpartum CVST is vital because it is associated with high mortality and morbidity. The diagnosis of CVST and multiple venous infarcts secondary to intracranial hypotension should be considered in the presence of a persistent postpartum headache following spinal anaesthesia/analgesia, especially if not postural. This case emphasises the importance of prompt diagnosis which may be rapidly cured with simple measures such as bed rest, hydration and anticoagulant.

Learning points

- It is important to consider intracranial hypotension and cerebral venous sinus thrombosis in the differential diagnosis of headache during the postpartum period in women with a history of prior spinal and/or epidural anaesthesia.
- There is a need for a multidisciplinary approach and the early involvement of neurologists and radiologists.
- Early use of correct diagnostic imaging could reduce morbidity and mortality associated with cerebral venous sinus thrombosis.
- Application of cranial MRI and MR venography together provide a correct and reliable diagnosis of nearly all patients with thrombosis or infarct.[11]

Competing interests None.

Patient consent Obtained.

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