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Case Report

CEREBRAL SINUS THROMBOSIS AFTER SPINAL ANAESTHESIA: PREDISPOSING FACTORS, OPERATING DIFFICULTIES AND DIAGNOSTIC PROBLEMS.

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ABSTRACT

The authors describe a case of cerebral venous sinus thrombosis following subarachnoid anaesthesia, characterized by operative difficulties and lack of patient collaboration.

After anaesthesia, the patient reported a positional headache and on the 5th day following surgery, he developed a frontal tensive headache with dysphoric and depressive symptoms. On the 9th day he presented with emesis, hypostenia of the upper left limb and ipsilateral positive Babinsky. The diagnosis was made only on the 10th day.

Genetic analyses showed a homozygous mutation of methylene-tetrahydrofolate-reductase associated to hyperhomocysteinemia (21,7μM).

The recurrence of post-dural puncture headaches is very frequent in patients undergoing subarachnoid anesthesia, but special attention is required in the event of headache, as well as associated clinical signs and other risk factors, in order to achieve a precocious diagnosis and allow the physician to start a suitable therapy.

1. Introduction

Thrombosis of the cerebral venous sinuses is a major acute, fortunately rare, cerebrovascular event, associated with many conditions. The etiology of this disease remains unknown [(1)].

Diagnosis of the disease is not always easy. As cerebral thrombosis can result in serious encephalic complications, early diagnosis and timely anticoagulant therapy, which will positively influence its clinical course, are essential.

2. Case presentation

Patient: male, aged 28, had undergone knee arthroscopy due to a meniscal lesion. Preoperative investigations (ECG, laboratory examinations and chest x-rays) did not show significant pathological abnormalities. Anesthesiologic risk was classified as ASA II, as the patient was a smoker and had mild hypertension.

The anesthesia technique adopted was a neuraxial subarachnoid block using hyperbaric 0.5% marcaine and a 22 gauche spinal needle. Due to operational difficulties and lack of patient collaboration, the anesthesiologist had to insert the needle at L2-L3 level several times before successfully performing the procedure.

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Premedication was given with atropine 0.3 + 0.3 mg iv boluses of, and a 500 ml preload of physiological solution. During surgery, 1,000 ml of physiological solution and 500 ml of 5% glucose solution were infused.

In the early postoperative period, the patient had a frontal headache with irradiation in the occipital region, increasing in its intensity on standing.

The next day, neurological examination did not detect pathological results, while psychiatric counseling showed a defective-depressive mood.

On the fifth day, the headache lost its postural characteristics and assumed a persistent fronto-orbital location. ORL consultation was also required due to a suspicion of frontal sinusitis.

On the seventh day, the patient was transferred to a neurological department; clinical examination was still negative for focal neurological signs, the headache was still present.

A CT scan showed "abnormal density" of the right transverse sinus, straight sinus and the back of the superior sagittal sinus.

On the ninth day, the patient manifested vomiting, psychomotor agitation, left side weakness and ipsilateral positive Babinski reflex.

A further CT scan showed a persisting density in the straight sinus and at the back of the superior sagittal sinus; coagulation tests showed a 100% prothrombin activity, 68% antitrombin III activity and a fibrinogen increase (514 mg%). The next day, the patient showed Jacksonian crises in the left hemisphere. A brain NMR was performed and revealed:

- Thrombosis of superior, straight and transverse sinuses.
- Small hemorrhage in right cerebellar hemisphere.
- Ischemic injury in the right temporal-parietal region associated with small hemorrhagic spots.
- Bi-hemisphere cerebral edema

The acquired diagnostic data resulted in the diagnosis of venous sinus thrombosis, so cortisone, mannitol and heparin therapy was initiated; antiepileptic drugs were also administered.

Subsequently, progressive remission of neurological deficits was observed and epileptic seizures were controlled sufficiently by pharmacological therapy; there was only a slight weakness in the left hemisoma.

Genetic examinations showed a homozygous mutation of the gene encoding the methylene tetrahydrofolate reductase enzyme, associated with an increase in serum homocysteine levels (21.μM).

The patient was discharged after twenty days of anticoagulant and antiepileptic therapy. Six months later, a RMN control of the brain showed a residual poroencephalic cavity in the right temporal lobe (Figure 1). At present, the subject in on antiplatelet (acetylsalicylic acid, 100mg / day) and antiepileptic (Gardenale 50 + 100mg / day) therapy; he still presents as significant clinical disorder in terms of a slight left-side weakness.

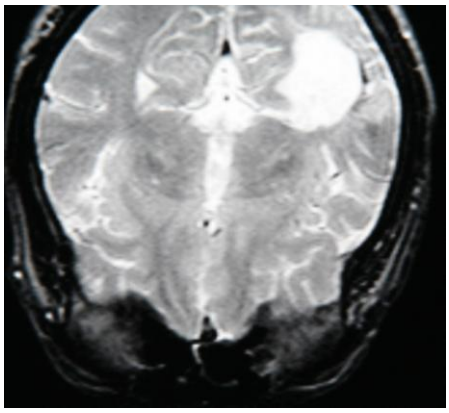

Figure 1 - Brain RMN performed six months after sinus thrombosis, with residual pore-brain cavity on the right temporal lobe.

3. Discussion

The Cerebral vein thrombosis (CVT) is a very rare disease, compared to the arterial occlusive diseases of the brain. It can cause serious neurological syndromes, especially between the ages of 20–35, and is responsible for 1–2% of all the strokes in adults. Predisposing factors are recognizable in 75% of cases (Table 1).

Local
• Trauma
• Infections
• Subarachnoid anaesthesia
Systemic
• Pregnancy and postpartum conditions
• Hypercoagulable states (Factor V Leiden mutation, Protein C and S deficiency, Anti-trombin III deficiency, Prothrombin gene mutation)
• Malignancy (leucemia, lymphoma)
• Oral contraceptive use
• Sickle cell anaemia
• Primary and secondary polycythemia
• Anti-phospholipid antibody syndrome
• Thrombocytosis
• Paroxysmal nocturnal haemoglobinuria
• Dehydration, hyperviscosity
• Rheumatic diseases (Behçet's disease, lupus, sarcoidosis, vasculitis, nephrotic syndrome)
• Cyanotic congenital heart disease
• Cachectic infants
• High body mass index
Idiopathic
•No reason in 25% of patients

Table 1 - Predisposing factors for CVT

Careful consideration of predisposing factors emphasizes the importance of coagulation disorders in the incidence of this disease. In fact, most of the cases described in the literature refer to pregnant or post-partum patients, occasionally undergoing neuraxial anesthesia, generally subarachnoid or epidural with possible accidental puncture of the dura. Venous thrombosis risk increases by 5–6 times in pregnancy and the incidence of thromboembolic disease related to pregnancy is 0.13%. In 50% of these cases, an hereditary pattern of thrombophilia, such as factor V Leiden mutation, prothrombin gene mutation, absence of anti-thrombin III, absence of protein C and/or protein S and hyperhomocysteinaemia, is found. In addition, dural puncture represents a trigger, for CVT.

In more detail, the literature reports cases of sagittal sinus thrombosis following subarachnoid anesthesia, especially in women undergoing subarachnoid analgesia in delivery[(2)]. The pathogenic mechanism described at the origin of this event refers to loss of cerebrospinal fluid, decreased cerebrospinal fluid pressure and intracranial hypotension, induced by subarachnoid anesthesia. The decrease of the cerebrospinal fluid pressure, together with its loss following dural puncture, may result in a stretching of the encephalic structures with potential damage to the venous endothelium. Anesthesia-induced vasodilation, consequent blood stasis and the patient's supine decubitus may induce the onset of cerebral venous thrombosis[(3)].

In addition, more extensive lesions, usually associated with execution errors of subarachnoid anesthesia, may cause a significant leakage of cerebral spinal fluid and contribute to the development of cerebral venous thrombotic phenomena (Table 2).

• Preexisting articular disorders
• Patient position
• Patient collaboration
• Seat of the puncture
• Needle type used
• Number of punctures
• Operator experience

Table 2 - Factors that can cause an excessive dural lesion with significant cerebrospinal fluid loss during subarachnoid anesthesia.

When the technique is properly performed, dural injury and spinocerebral fluid loss are not significant, and do not induce any particular symptoms, but they may sometimes cause a “typical” post-dural puncture headache (PDPH).

In the present case, the chosen device seemed to have an excessive caliber (22 G) in relation to the age and the expected difficulty of execution; furthermore, the patient underwent multiple attempts during the anesthesiological procedure: the needle was repeatedly reinserted before reaching the subarachnoid space, setting the conditions for multiple dural punctures and consequent significant loss of cerebrospinal fluid.

These technical difficulties during anesthesia, by increasing the cerebrospinal fluid loss and the subsequent endocranial hypotension, almost certainly imply a correlation between this procedure and thrombosis of the cerebral venous sinuses.

However, a key element in the case was the discovery of the patient’s clotting disorder; he was also affected by a homozygous mutation of the gene encoding for the methylene-tetrahydrofolate reductase enzyme associated with an increase in serum homocysteine levels (21.7 μM) and a mild reduction of antithrombin III (68% activity).

In summary, in the observed case, the technical and operational difficulties acted as trigger factors on a predisposed subject; the presence of thrombophilic factors, determining thrombosis of the cerebral venous sinuses.

However, diagnosis was achieved on the tenth postoperative day, only when a widespread thrombosis of the venous sinuses had already occurred, inducing a cerebral ischemic injury. Indeed, the classic symptoms associated with thrombosis of the venous sinuses, their clinical onset and course are variable, ranging from a simple headache to severe alterations of consciousness and behavior.

Headache, which is the most frequent and early symptom, is associated with various neurological symptoms in 90% of the cases (Table 3).

1. Endocranial hypertension syndrome
2. Focal neurological disorders
3. Alterations of consciousness and/or behavior
4. Cavernous sinus syndrome

Table 3 - Clinical manifestations related to cerebral venous sinus thrombosis.

The broad spectrum and the lack of specificity of clinical manifestations of venous thrombosis make early diagnosis, which is useful to prevent major complications, extremely difficult.. In addition, the International Headache Society’s (IHS) Guidelines are not particularly helpful in correlating the headache to a cerebral sinus thrombosis[(4)].

As in most cases, the first clinical manifestation is the headache only, a relatively common symptom after spinal anesthesia, this symptom generally does not lead to diagnosis, as long as the clinical findings do not reveal any further symptoms. Consistently, Schievink[(5)], , found that 17 of 18 patients with spontaneous endocranial hypotension initially had an erroneous diagnosis, but, on the other hand, many other authors have emphasized the need to pay special attention to cases where the typical headache following spinal puncture, loses its postural characteristic, becoming persistent and worsening[(6)].

A further element of doubt should be the very early onset of the postural headache, , compared with the timing of the anesthesiological procedure, considering that in PDPH syndrome the onset of the headache is related to the rate of loss of spinocerebral fluid and posture, in this case clinistatic in bed, taken by the patient in the immediate postoperative period.

On the 3rd day, the headache was associated with behavioral alterations and, on the fifth day, lost its postural features to assume a fronto-orbital localization; on the seventh day the TC scan showed a mild hyperdensity of the meningeal sinuses.

The diagnosis of thrombosis of the cerebral sinuses was carried out only on the tenth day, when the patient presented Jacksonian crises and the brain MRI showed the straight, transverse and sagittal sinus thrombosis as well as a "hemorrhagic area at the right temporo-parietal lobe and bilateral cerebral edema", in association with laboratory data of thrombophilic alteration.

Radiological signs are not always specific. Digital angiography represents the reference diagnostic procedure, and the contrast enchanced MRA (MRI) can provide similar diagnostic results.

4. Conclusions

Spinal anesthesia represents an invasive procedure that can determine a severe event, as a result of drugaction or some other pathology [7-10].

The atypical features, even though minimal, of an headache and its tendency to change, observed in a patient who has undergone spinal anesthesia, especially when associated with difficulty of execution, require more diagnostic attention, and would encourage more extensive clinical and instrumental examinations (fundus oculi, TC scan, angiography, etc) able to reveal a venous thrombotic process.

References

1. Appenzeller S, Zeller CB, Annichino-Bizzacchi JM, Costallat LT, Deus –Silva L, Voetsch B, Faria AV, Zanardi VA, Damasceno BP, Cendes F. Cerebral venous thrombosis: influence of risk factors and imaging findings on prognosis. Clin Neurol Neurosurg, 2005;107:371-8.
2. Dadheech R, Khandelwal M, Chauchan S, Sharma SP. A case of postpartum lateral sinus thrombosis following cesarean section under spinal anesthesia. J Anaesthesiol Clin Pharmacol, 2016;32:274-5.
3. Mahesh PK, Bejoy T, Sylaja PN. Cerebral venous thrombosis in post-lumbar puncture intracranial hypotension: case report and review of literature. 2014, Version 1. F1000Res. 3:41.
4. Headache classification committee of the HIS Classification and diagnostic criteria for headache disorders cranial neuralgias and facial pain, Cephalagia, 2004, 24(supl. 1).
5. Schievink WI. Misdiagnosis of spontaneous intracranial hypotension. Arch Neurol, 2003;60:1713-8.
6. Todorov L, Laurito CE, Schwartz DE. Postural headache in the presence of cerebral venous sinus thrombosis. Anesth Analg, 2005;101:1499-1500.
7. Massoni F, Ricci P, Simeone C, Ricci S. Cardiac death in aortic valve sclerosis and coronary artery disease. An autopsy report . Act Med Med 2014;30(1):77-80.
8. Archer T, Ricci S, Garcia D, Ricciardi MR. Neurodegenerative aspects in vulnerability to schizophrenia spectrum disorders. Neurotox Res. 2014;26(4):400-13.
9. Vitarelli A, Martino F, Capotosto L, Martino E, Colantoni C, Ashurov R, Ricci S, Conde Y, Maramao F, Vitarelli M, De Chiara S, Zanoni C. Early myocardial deformation changes in hypercholesterolemic and obese children and adolescents: a 2D and 3D speckle tracking echocardiography study. Medicine (Baltimore). 2014;93(12):e71.
10. Vyshka G, Vacchiano G. Severe flaccid paraparesis following spinal anesthesia: a sine materia occurence. BMJ Case Report, 2014, Vol. 5, pii: bcr2013202071