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CASE REPORT

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Life-threatening intracranial hypotension after diagnostic lumbar puncture

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Abstract Intracranial hypotension syndrome as a complication of diagnostic lumbar puncture is a rarely observed entity. Intracranial hypotension syndrome is characterized by postural headache, neck pain/stiffness, blurred vision, nausea, vomiting, clouding of consciousness, dizziness and vertigo. The majority of cases resolve spontaneously with conservative treatment. Rarely, epidural blood patch is required. We report a 41-year-old man with multiple sclerosis, who developed intracranial hypotension syndrome after diagnostic lumbar puncture and who did not respond to conservative treatment. A subdural hematoma was subsequently found, when the patient showed considerable worsening of clinical conditions with life-threatening symptoms. Surgical evacuation of the subdural hematoma was not sufficient to improve significantly the patient's conditions, while complete symptoms remission was achieved 12 hours after epidural blood patch. We stress the need for epidural blood patch in any case of post-diagnostic lumbar puncture postural headache which does not resolve with conservative therapy.

Key words Intracranial hypotension syndrome • Epidural blood patch • Diagnostic lumbar puncture • Subdural hematoma • Postural headache

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Introduction

Intracranial hypotension syndrome (IHS) was first described by Schaltenbrand in 1938 as a cause of postural headache [1]. This author defined the syndrome as "spontaneous aliquorrhea" and proposed three mechanisms to explain low cerebrospinal fluid (CSF) pressure: (a) diminished CSF production; (b) hyperabsorption of CSF; and (c) CSF leakage through small meningeal lacerations. Today, most authors agree that etiology is related to meningeal tears, intentional or accidental; IHS may be caused through diagnostic lumbar puncture (DLP), myelography, spinal anesthesia, spinal injury and spinal surgery [2–9]. Several authors reported cases in which spontaneous spinal CSF leakage through congenital meningeal fistulas resulted in the characteristic postural headache of IHS (primary or spontaneous forms) [10, 11].

Postural headache represents the most characteristic symptom of IHS; it usually begins in the frontal region or vertex, but may also be suboccipital. Pain is dull, deep, and aching, at times throbbing. Although the exact pathogenic mechanisms remain unclear, postural headache is thought to result from continuous CSF loss through the dural hole. CSF loss interferes with the normal cushioning of the intracranial contents. As a result, intracranial contents may descend when the patient is standing upright, leading to traction on pain-sensitive structures in the dura mater [7, 9, 12, 13]. Other clinical findings in patients with IHS include symptoms such as diplopia, photophobia, nausea, hyperacusis, vomiting, bradycardia, nystagmus, abducens nerve palsy, binasal visual field restriction, neck stiffness, dizziness, tin-

nitus, vertigo and disturbances of equilibrium. Mental symptoms may occur as well, including clouding of consciousness, mental impairment, and drowsiness [4].

A characteristic constellation of findings on magnetic resonance imaging (MRI) has been described in IHS patients [5, 11, 14–16]: (a) images show diffuse meningeal enhancement after gadolinium administration, a symmetrical thickening which is often referred to as pachymeningitis; (b) axial images show effacement or crowding of the sulci, gyri, Sylvian fissures, and basal cisterns; (c) mid-sagittal images show generalized descent of midline structures that crowd the posterior fossa. The thalamus is displaced partly through the incisura, the cerebral aqueduct descends into the posterior fossa, the pons is deformed against the clivus, and there is tonsillar herniation; and (d) last, subdural hygromas frequently develop and occasionally progress to form secondary hemorrhages [5, 7, 8, 12, 17, 18, 19, 20]. At liquoral manometry, low opening pressure confirms IHS [2, 6, 7, 12, 21].

Although lumbar puncture is generally well tolerated, the occurrence of post-dural puncture headache can be one of the more frequent adverse effects associated with this procedure [22–25]; however, the majority of cases will resolve spontaneously in 3–7 days with conservative treatment. We have been able to identify only four cases of severe and persistent IHS induced by diagnostic lumbar puncture; one was fatal, due to tonsillar herniation [26], while the other three patients developed subdural hematomas [8, 12, 26]. In these last cases, surgical evacuation was sufficient to resolve the situation, differently from our case, in which surgical drainage proved not to be able to resolve it, thereby rendering necessary an epidural blood patch (EBP), which succeeded fully. EBP produces complete resolution of headache in 70% of patients. Thus, EBP is effective in IHS therapy and a relapse after treatment does not always require a second patch. However, medical specialists other than those in anaesthesia are reluctant to accept the benefits of EBP.

Case report

A 41-year-old man was admitted to our department because he had developed progressive, bilateral lower limb motor impairment since the age of 39 years. He reported a past episode of double vision and dizziness at age 24 years. Neurological examination revealed a pyramidal syndrome affecting all extremities. He was hospitalized and subjected to visual (VEP) and somatosensory (SEP) evoked potentials which pointed to central pathway involvement. To confirm the suspicion of multiple sclerosis, lumbar puncture was performed in clinostatism on left side, using a 20-gauge needle, and 5 ml CSF was withdrawn. Immunoelectrophoresis showed intrathecal IgG synthesis, thereby confirming the diagnosis of multiple sclerosis.

After two days the patient was discharged. After five days he was rehospitalized due to the development of persistent postural headache, mainly in the frontal region, which could be attenuated only by lying down. For about 3 days, the patient was maintained in the lying position as the upright position immediately exacerbated his headache.

Computed tomography (CT) showed the existence of a limited left frontoparietal subdural collection. As the clinical picture started to deteriorate rapidly, with the onset of an altered state of consciousness, the patient underwent emergency MRI, which showed the existence of a left subdural hematoma compressing the homolateral cerebral hemisphere and dislocating and deforming the corresponding cerebral peduncle subtentorially, as evidenced on T1-weighted coronal images (Fig. 1). Sagittal T1-weighted images showed cerebellar tonsil descent in the foramen with brainstem compression and disappearance of the cisterna magna (Fig. 2).

The patient was immediately transferred to the operating room for evacuation of the subdural hematoma. During transfer, his conditions worsened further with the onset of Cheynes-Stokes' respiration. At operation, the dura mater appeared to be detached from the skull and the hematoma was not hypertensive. A subdural probe for continuous monitoring of intracranial pressure (ICP) was applied. After drainage, the patient's conditions improved only mildly, with the relief of Cheynes-Stokes' respiration. Since ICP yielded values of 0–5 mmHg, it was decided to perform an epidural blood patch in the lumbar region with 20 ml autologous blood. Within 12 h, the patient's neurological picture resolved completely; he was discharged after four days. A control MRI examination performed at discharge showed complete normalization of brain structures (Fig. 3).

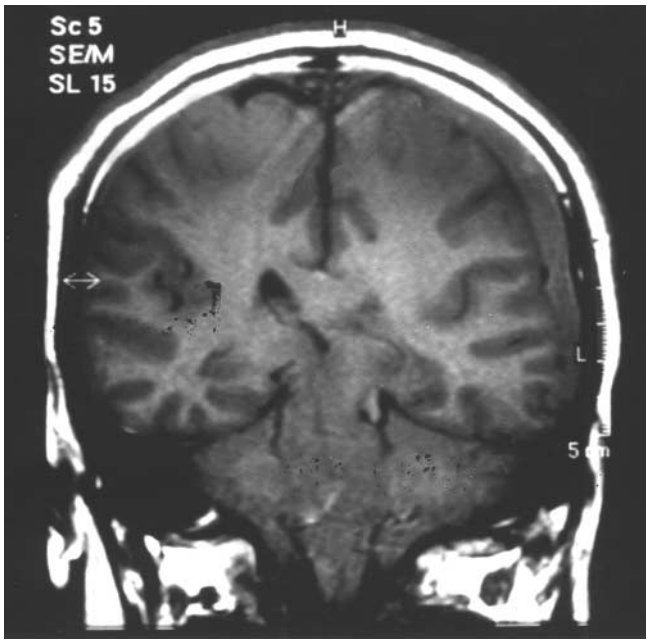

Fig. 1 T1-weighted coronal MR image performed 10 days after diagnostic lumbar puncture, showing the existence of subdural hematoma compressing the left cerebral hemisphere and dislocating and deforming the corresponding cerebral peduncle subtentorially. Note the failure to visualize the left lateral ventricle

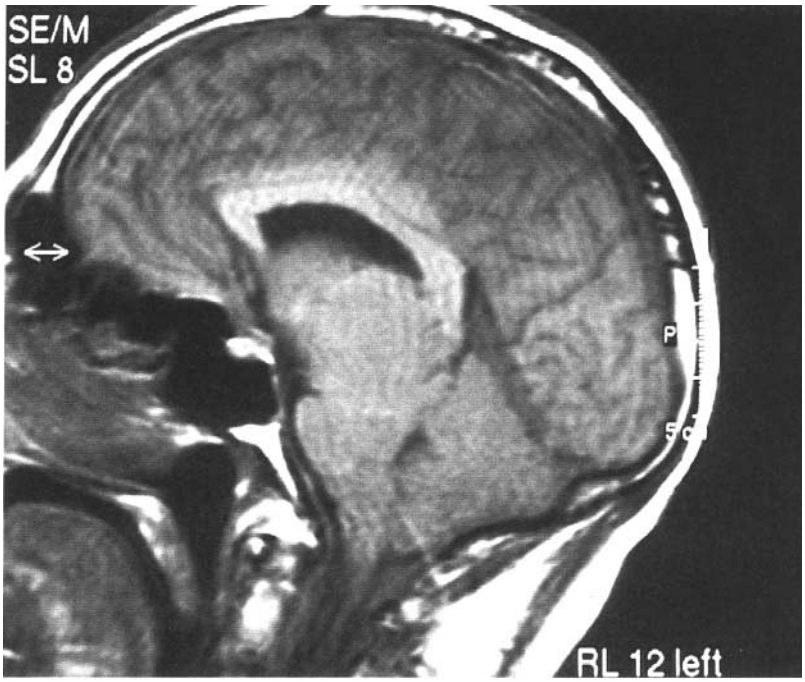

Fig. 2 T1-weighted sagittal MR image performed after subdural hematoma drainage, showing cerebellar tonsil descent within the foramen with brainstem compression, as well as prepontine cisterna and cisterna magna disappearance

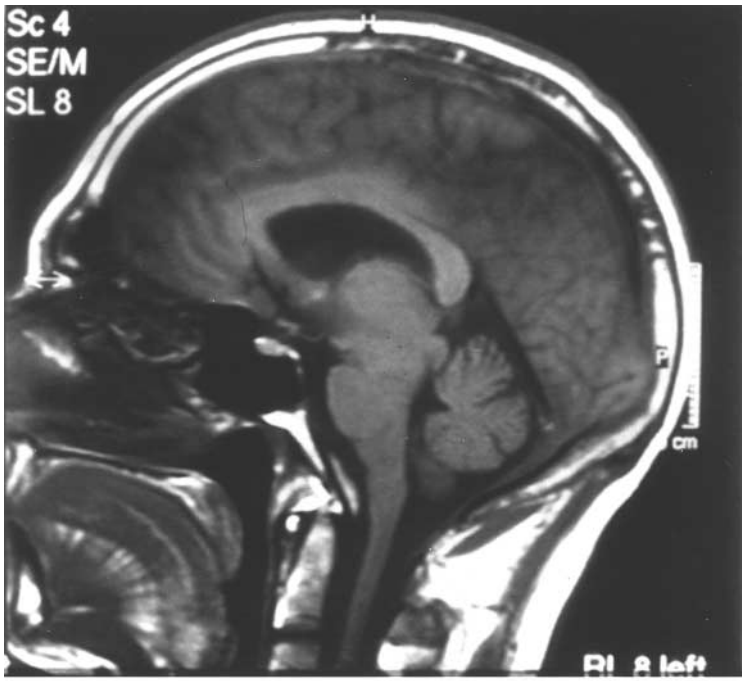

Fig. 3 Control T1-weighted sagittal MR image performed at discharge, showing complete normalization of brain structures: cerebellar tonsil ascent, no evidence of brainstem compression and reappearance of the cisternas magna and prepontine

Four months later, the patient was well. He had resumed his everyday life activities, was doing well at job and the symptoms of multiple sclerosis had improved.

Discussion

Postural headache is a typical and pathognomonic symptom of intracranial hypotension caused by the stretching of meningeal pain-sensitive terminals. It is a common sequela of DLP (occurring in 30%–40% of cases), and most often it resolves either spontaneously or with analgesics and best rest. Other accompanying signs or symptoms are also related to liquoral hypotension and to caudal displacement of brain structures, with consequent loss of the pressure effect of CSF, hence traction and distension of central and peripheral sensorimotor structures of the neuraxis [7, 9, 12, 13]. Intracranial hypotension, whatever its cause, is associated with subdural hematoma in 10% of cases [5, 7, 8, 12, 19, 20,

26]. To date, 50 cases of severe IHS with subdural hematoma resulting from spinal anesthesia, accidental dural laceration, myelography, intrathecal chemotherapy, or diagnostic lumbar puncture have been reported in the literature.

Our case is characterized by the onset of post-DLP headache which did not respond to conservative treatment. DLP was performed in our patient to confirm diagnosis of multiple sclerosis. The fact that we used a large caliber needle (20 G) could have increased the risk for IHS [8, 9, 12, 22, 23, 27–31]. Five days after discharge, the patient had to be rehospitalized due to postural headache, and a CT scan, immediately performed at rehospitalization, showed the existence of a left frontoparietal subdural hematoma of limited extension. As the patient's clinical conditions were rapidly deteriorating and MRI showed a downward brain shift, the hematoma was evacuated; an ICP monitoring probe was placed. Since the pressure was low and the patient's conditions did not significantly improve after the operation, a lumbar EBP was performed. This was followed by immediate resolution of symptoms in 12 h.

In the anesthesia literature, it is strongly recommended that patients be treated with EBP from 5 to 7 days after the onset of persistent headache, independently from whether it is postural or not [27–33]. It might have been appropriate to have treated the patient with EBP earlier, as soon as hematoma was found. We suppose that such treatment could have resolved the clinical picture before the development of life-threatening symptoms, rendering the hematoma drainage unnecessary.

We suggest that when postural headache lasts for more than one week and other diagnoses have been ruled out, treatment with EBP ought to be considered. If DLP is to be performed, it is preferable to use a small caliber needle to decrease the risk for IHS. We stress the need for EBP in any case of postural headache which does not resolve with conservative therapy; its clinical utility has been probably underestimated to date.

Sommario *La sindrome da ipotensione liquorale (IHS) può essere raramente la conseguenza di una puntura lombare eseguita a scopo diagnostico. Essa è caratterizzata da cefalea posturale, dolore o rigidità nucale, visione offuscata, nausea, vomito, vertigini e stato confusionale. Nella maggior parte dei casi la IHS tende alla remissione spontanea con terapia conservativa. Raramente si rende necessaria la terapia chirurgica mediante "patch" epidurale (*epidural blood patch*, EBP). Descriviamo il caso di un paziente di 41 anni affetto da sclerosi multipla che, dopo rachicentesi lombare effettuata a scopo diagnostico, ha sviluppato una IHS non responsiva alla terapia medica. Successivamente, ha sviluppato un ematoma subdurale che si accompagnava ad un peggioramento delle condizioni cliniche generali e neurologiche fino a sfociare in una prognosi riservata* quoad vitam. *L'evacuazione dell'ematoma non ha modificato le condizioni cliniche mentre abbiamo assistito alla remissione completa della sintomatologia poche ore dopo la esecuzione di un EBP. Vogliamo focalizzare l'attenzione sulla opportunità di eseguire un EBP in tutti i casi di IHS che risultano resistenti alla terapia conservativa.*

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