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Cranial nerve VI palsy and spontaneous intracranial hypotension

Parálisis del VI par craneal e hipotensión intracraneal espontánea

Dear Editor:

We report the case of a 40-year-old woman, with no personal history of interest, with a clinical picture of a 15-day frontal and occipital headache of oppressive evolution. It worsened with sitting and improved with analgesia and in supine position. Subsequently, she began to suffer from photophobia and diplopia. There was no history of trauma.

The general examination was unremarkable and vital signs were normal. The neurological examination highlighted a palsy of the left sixth nerve with horizontal diplopia to the left, which disappeared when looking to the right; there were no other abnormalities.

Diagnostic tests showed no abnormalities in chest x-ray, electrocardiogram, cranial computed tomography (CT), general analysis, microbiological culture, serology (syphilis, *Borrelia*, HIV and *Brucella*), angiotensin converting enzyme (ACE), adenosine deaminase (ADA), and cerebrospinal fluid (CSF) anatomopathology.

Cranial magnetic resonance imaging (MRI) with paramagnetic contrast cranial (Fig. 1) showed a thickening and intense leptomeningeal enhancement in the absence of subdural collections or tonsillar descent; the rest of the examination was normal. The lumbar puncture (LP) showed an opening pressure of 3 cm H_2O, clear CSF, glucose at 83 mg/dl and protein at 158.1 mg/dL; 28 erythrocytes/μl and 8 leucocytes/μl. The isotopic cisternoscintigraphy (Fig. 2) performed showed an image indicative of CSF leakage in the paraspinal region at the level of the cervicothoracic junction. The study was completed with a cervicothoracic MRI (Fig. 3), which showed an image of subdural CSF collection from C6 to D3, compatible with CSF fistula at the cervical level. The correction of physiological cervical lordosis and posterior osteophytosis with accompanying disk in the C5-C6 space indenting the anterior epidural space could also be appreciated.

Conservative measures were initiated due to the liquoral hypotension (abundant hydration, relative rest, analgesia and corticosteroids). When these proved ineffective, an autologous epidural blood patch at the lumbar level (20 ml) was administered; a progressive improvement in headache (70-80% improvement) and diplopia (30% improvement) took place after one week. Within a month, the headache had disappeared and a lower limitation of the abduction of the sixth cranial nerve was observed, with diplopia only in extreme lateral gaze to the left, which disappeared in subsequent months. After 4 months, spinal MRI showed no CSF collection, confirming the absence of CSF leakage, as could be expected from the clinical evolution.

Intracranial hypotension syndrome (IHS) is secondary to a depletion of CSF volume. CSF leakage is postulated as one of the major aetiopathological causes, producing a rostrocaudal descent of the brain, pulling on pain-sensitive structures, cervical roots and the oculomotor nerves. The causes are often iatrogenic (lumbar puncture, spinal anaesthesia, myelography, massive CSF drainages, cranial or spinal surgery) or secondary to trauma. It may also be observed spontaneously, from trauma or banal efforts that

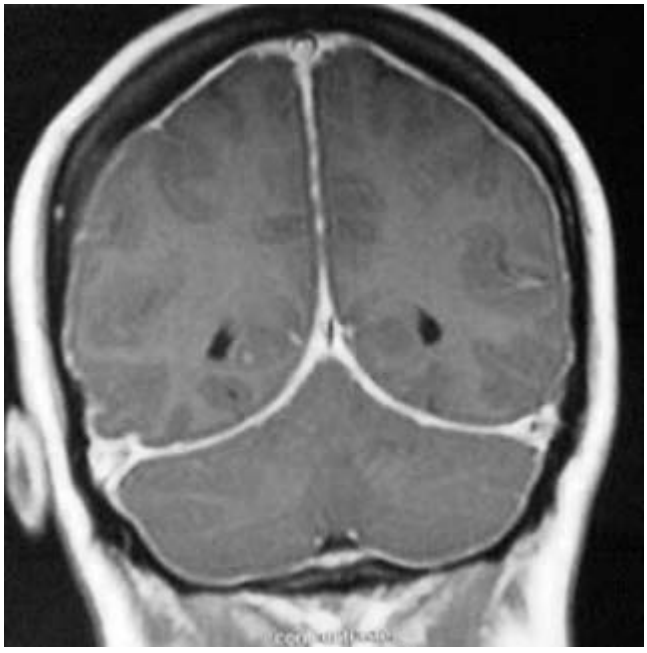

Figure 1 Study with cranial magnetic resonance imaging after administration of paramagnetic contrast, T1 sequence. Marked leptomeningeal uptake can be observed.

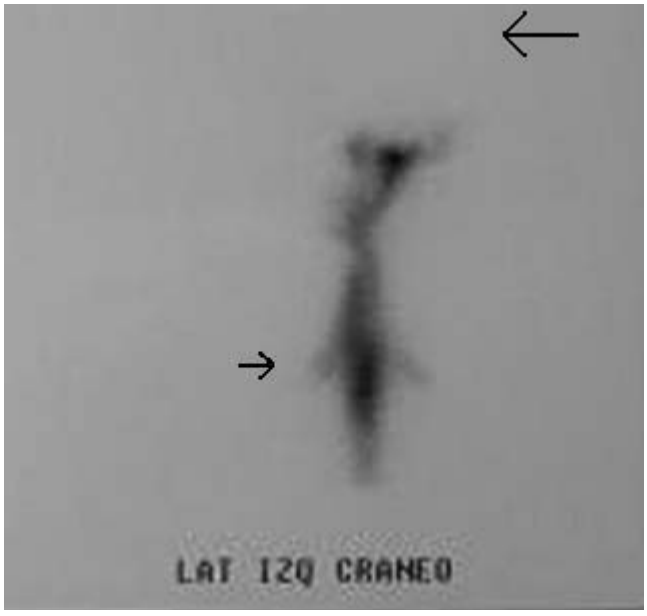

Figure 2 Cisternoscintigraphy with radiopharmaceutical ^{111}In-DTPA administered intrathecally. The presence of two bands of activity in a paraspinal location at the cervicothoracic junction can be observed, indicating a cerebrospinal fluid leakage (short arrow). There is no identification of passage of the radiopharmaceutical to the cerebral convexity after 24 and 48 h (long arrow).

produce small dural lacerations. Clinically, it is characterised by headache that appears or worsens when moving from a supine to a standing position and which disappears or

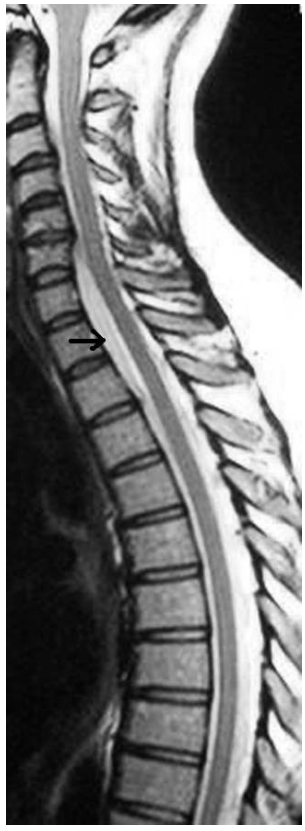

Figure 3 Magnetic resonance imaging study after administration of paramagnetic contrast in the cervical and thoracic spine, T2 sequence. It shows leptomeningeal collection at the cervical and thoracic levels and an image indicative of cerebrospinal fluid collection in the anterior epidural space from C6 to D3 (arrow).

decreases when lying down. It can be associated with nausea, vomiting, cervical pain, interscapular pain, dizziness, diplopia (in up to 30-35% of cases[1]), blurred vision, transient visual obscuration, visual field defects, hearing disorders and radicular symptoms[2,3].

Our patient presented a spontaneous IHS, in which CSF leakage could be detected in the cisternography at the cervicothoracic level by an image of subdural CSF collection from C6 to D3 in the spinal MRI. The association between disc protrusions and CSF leakages had already been described, indicating the possible origin of the dural laceration in that area[3]. The diagnosis of spontaneous HIS in one of the most common locations was thus confirmed, with progressive clinical recovery after administration of an epidural blood patch. This treatment is effective in hypotension secondary to lumbar puncture. In spontaneous cases, the evidence comes from case series, indicating a positive effect not without risks[1,4-8]. In this case, the benefit was complete, with total symptom resolution and absence of CSF collection in the spinal MRI after treatment.

It is important to bear in mind that the combination of spontaneous intracranial hypotension and oculomotor disorders[5] is frequent, so this entity has to be taken into

account in cases of headache and diplopia. Despite the absence of placebo-controlled studies, given the proven efficacy in case series, we believe that treatment with epidural blood patches should be considered in these cases.

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