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CASE REPORT

A case of late orthostatic headache secondary to lumbar hidden dural fistula 20 months after epidural anesthesia

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Key Clinical Message

The clinical recognition of CSF fistula is a clinical challenge. We report the case of a young woman, who presented with a late orthostatic headache 20 months after epidural anesthesia. She developed a lumbar dural fistula of CSF confirmed in myelography CT scanning and treated successfully with epidural blood patch.

Keywords

CSF fistula, epidural anesthesia, epidural blood patch, headache unusual appearance.

Introduction

Cerebrospinal fluid (CSF) fistula is a recognized cause of intracranial hypotension and a rare complication of procedures accessing the neuraxis. The clinical recognition of this entity is a challenge for the clinician. We describe a case of late orthostatic headache 20 months after epidural anesthesia that caused a lumbar dural fistula of CSF, confirmed in myelography computerized tomography (CT) scanning and treated with epidural blood patch (EPB) with autologous blood leading to complete disappearance of her symptoms.

Case Report

We describe the case of a 36-year-old Caucasian woman, store-housekeeper store who, 4 days before admission (on 19 December 2015), as she was driving, was struck by an intense and oppressive left front-parietal headache, without maximum intensity from onset, accompanied by photophobia, repeated vomiting, and tinnitus in the left ear, without autonomic complaints. Our patient felt worse when standing and relieved after 10 min of supine position, with no improvement with paracetamol. There was no personal or family history of headache. She had a healthy lifestyle and was not overweight.

In her past medical history, she had recurrent otitis in childhood, habitual hypotension and an episode of lumbar-sciatica pain with irradiation to the left leg several years ago and sporadic low back pain on effort, without reported headache. With regard to anesthetic or surgical history (outside our hospital), she had an excision of a cyst on the right foot with general anesthesia in 2002; in 2007, she underwent a cesarean under general anesthesia;

in 2010, she had a cesarean section with epidural anesthesia in a sitting position with use of catheter for postoperative analgesia, and in 2014 (20 months prior), she sustained a cesarean section with epidural anesthesia (18-G Tuohy needle) at the L4-L5 level, in the sitting position. In all anesthetic procedures, there were no incidents manifested by the patient nor registered in the clinical records.

In the emergency room on admission, the general and neurological examinations were normal. The blood pressure was 94/58 mm Hg, the pulse was regular and at 100 beats per minute, and the ECG was in sinus rhythm. The cranial CT was normal. She was admitted to the Neurology Service for further investigations with the aim of finding a dural fistula as the first diagnostic hypothesis because it was always a spontaneous orthostatic headache accompanied by tinnitus without meningism. She performed a brain MRI with contrast and MR angiography, which showed inflammatory hypertrophy of the mucosa of the maxillary sinuses, without subdural fluid collections, enhancement of the pachymeninges, or sagging of the brain with good positioning of the cerebellar tonsils. Other causes of orthostatic headache were excluded as a colloid cyst of the third ventricle, the Chiari malformation and the postural orthostatic tachycardia syndrome. She was examined by otorhinolaryngology, and there were no alterations, including the audiogram. Blood test was normal (C-reactive protein, complete cell count, prothrombin time, partial thromboplastin time, INR, electrolytes, folate, vitamin B12, thyroid and lipid profiles, uric acid, autoimmune tests and VDRL, hepatitis, and HIV-1 and HIV-2 antibodies).

She began symptomatic treatment. As symptoms persisted in the third week of admission, a neuraxial MRI was performed (Fig. 1A) with suspected dural CSF fistula, showing liquor signal distal to the dural sac, on STIR sequence. As it was not possible to detect the exact point of fistula with MRI, we performed myelography CT scanning (Fig. 1B–E) that confirmed the lumbar dural fistula of CSF, by best detection of distal extradural and extracanalar iodine contrast, also not showing the exact location of the fistula. Nevertheless, it oriented blood patch to the lumbar vertebral segment.

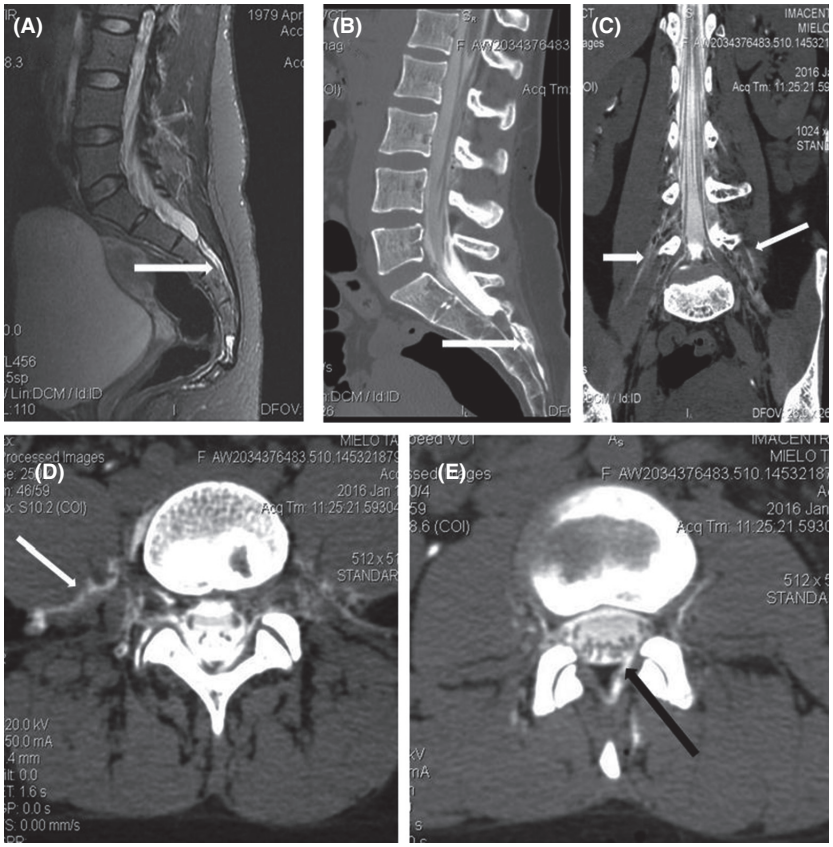

Figure 1. Short T1 Inversion Recovery (STIR) MRI (A): Sagital (A), arrows indicate CSF signal outside the dural sac, an indirect sign of dural fistula. Myelography CT scanning (B–E): sagital (B), coronal (C), axial (D, E). Arrows show extradural and extracanalar contrast, by extravasation of CSF. At L3 level (E), the site of dural puncture for myelography CT scanning.

On 22 January 2016, support from the Pain Unit was requested, and the patient underwent epidural saline patch (test). 10 cc. saline to 0.9% was injected at the L3-L4 level in single shot, followed by continuous infusion through epidural catheter of saline at 10 mL/h for 8 h, 5 mL/h for 12 h, and 2 mL/h for 48 h. As this test was positive, and after 24 h of complete rest, she began walking without orthostatic headache; finally, on the second of February 2016, she was submitted to an EBP at the L2-L3 level, in the left lateral decubitus with 25 mL of autologous blood extracted under strict aseptic technique. After 24 h of rest, she stopped having headaches in orthostatism, reporting only to back pain of mild intensity without radicular irradiation and no signs of meningeal irritation. She was discharged on the sixth day after the intervention without any complaints. She was advised some rest and low professional activity. Three weeks after discharge, she was observed at the ambulatory care unit showing neither orthostatic headache nor back pain. The general and neurological examinations were normal. A month later, she reinitiated her professional activity.

Discussion

Our patient had a headache secondary to dural fistula, typically an orthostatic headache that it was soothed by lying flat. No CSF pressure was measured because it could worsen the symptoms. Despite previous anesthetic procedures, symptoms appeared 20 months after the last epidural anesthesia. Thus, we thought this might be the causal antecedent of the symptoms. This latter description is a rare presentation of postdural puncture headache, probably accidental and unexpected. According to the International Classification of Headache Disorders in its 3rd edition, our case meets the criteria for CSF fistula headache [1] (A. Any headache fulfilling criterion C. B (1) A spinal procedure has been performed, or trauma has occurred, know sometimes to cause persistent CSF leakage (CSF fistula). (2) Low CSF pressure and/or evidence of low CSF pressure and/or of CSF leakage on MRI, myelography, CT myelography, or radionuclide cisternography. C. Headache has developed in temporal relationship to the procedure or trauma. D. Not better accounted for by another ICHD-3 diagnosis).

Younger women may be at a greater risk because of increased dural fiber elasticity that maintains a patent dural defect compared to a less elastic dura in older patients [2].

In our case, although the underlying pathophysiology is uncertain, we thought a fragile area at the puncture site might have played a role and, for example, by straining (Valsalva and other factors), an area of the dura had been torn and/or the loss of CSF might have caused compensatory venodilation to intracranial hypotension, which might have produced the headache [3]. For some authors [4, 5], the location of the catheter in the vicinity of the place where there was puncturing of the dura may predispose to the formation of CSF fistula [6].

In conclusion, in the presence of a persistent orthostatic headache without clear time relationship with lumbar puncture and with normal brain MRI, it is necessary to embark on extensive search for a dural fistula in the neuraxis [7]. Finally, EBP is the treatment of choice when conservative treatment fails.

For all we know, we did not find in the medical literature any case of headache postlumbar dural fistula of CSF of installation so late after anesthetic procedure [8].

Authorship

FJRF: is the responsible doctor and corresponding author, has developed the text and the discussion. MADC: has contributed by pain history, has performed the EPB and helping with the discussion. RPP: has contributed by providing the images and helping with the discussion.

Conflict of Interest

The authors declare that there are no conflict of interests.

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