LETTER TO THE EDITOR

To The Editor

Acephalgic Intracranial Hypotension: Subdural Haematoma Resolution After Blood Patch

Keywords: intracranial hypotension, epidural blood patch, spontaneous intracranial hypotension, SIH, spreading cortical depression, spinal meningeal cyst, tarlov cyst

A cognitively unimpaired 73-year-old female was transferred to our centre following increasingly frequent spells of right hemiparesis and aphasia. These episodes started 2 months prior to her transfer and consisted of stereotypical and progressively increasing symptoms that reached nadir in a few minutes before improving spontaneously. The patient also complained of intermittent visual hallucinations that sometimes accompanied the paretic episodes. These spells had occurred a dozen times, with increasing frequency. In addition, in the few weeks preceding the patient's transfer to our centre, she developed psychomotor slowing, being unable to take part in meaningful conversations with her family and she was disoriented at times. On imaging, there were bilateral hygromas that progressed then complicated by acute bilateral subdural haematomas (SDH) (Figure 1(A)). The referring centre initiated levetiracetam that was titrated up to 1000 mg BID, without significant improvement. Relevant past medical history includes stable rheumatoid arthritis (RA) and pulmonary fibrosis due to methotrexate toxicity. There was no history of trauma and the patient had no headaches, in particular no orthostatic headaches.

Physical examination was normal, except for psychomotor slowing. A 20-minute electroencephalogram (EEG) was normal, without focal slowing or epileptiform discharge. Brain magnetic resonance imaging (MRI) with gadolinium revealed smooth pachymeningeal enhancement, in addition to the bilateral SDHs (Figure 2(A)). No lumbar puncture was performed prior to this study. The radiological findings were highly suggestive of spontaneous intracranial hypotension (SIH). A spine MRI with gadolinium was then performed and showed multiple meningeal cysts at sacral, lumbar, dorsal and cervical levels (Figure 2(B)). The largest cysts were at the sacral levels. No cerebrospinal fluid (CSF) leak was found.

Neurosurgical and spine surgery consultations were sought and concluded that no surgical intervention was indicated at that time, for both the SDHs and the spinal cysts. The patient's spells were attributed to cortical spreading depression caused by SDHs, and her treatment was changed from levetiracetam to topiramate. Given the hypothesis that there was an unidentified CSF leak that led to SIH and subsequent SDH formation, an epidural blood patch (EBP) was performed at L1–L2 with 15 ml of autologous blood. The patient had no paretic, dysarthric or visual spell during her 5-day hospitalisation on our ward. She significantly improved her cognitive status and was discharged home.

On follow-up 2 months after, the patient had no recurrence of neurological spells and was back to her prior functioning level. A head CT was done 3 months after her discharge. It showed complete resolution of the left hemispheric SDH and marked reduction in the size of the right SDH (Figure 1(B)). There was also no clinical evidence of rebound increased intracranial pressure. Altogether, follow-up was in favour of a benefit of EBP on the SIH and contributed to the resolution of the paretic spells and of the SDHs that had been progressively increasing in size for 2 months prior to the intervention.

Intracranial hypotension typically results from a CSF leak, which is almost always due to a traumatic or spontaneous spinal leak of various aetiologies.[1,2] Spinal meningeal cysts are fluid-filled diverticula of the spinal meningeal sac, nerve root sheath or arachnoid.[3] They are highly associated with SIH, although often incidentally found in healthy individuals.[4] In a well-conducted retrospective study on 568 patients with SIH referred to a tertiary care centre, 43% had meningeal cysts, with 22% of them showing an active CSF leak.[5]

The interest in our case lies in the fact that the patient was completely acephalgic, hence not meeting the International Classification of Headache Disorders (ICHD) criteria for SIH. No traumatic event was found on a careful history to justify an alternative cause of SDHs and of a presumed CSF leak. However, the presence of dural enhancement and the numerous meningeal cysts put SIH as the mechanism sustaining the SDHs. SDH is a well-documented complication of subdural hygroma related to SIH.[2] Pathophysiologically, in SIH, lack of CSF volume and pressure cause traction on the sensory nerves of the meninges and on the bridging veins; the latter being thought to cause SDHs.

Several case series report the safety and effectiveness of EBP in patients with SIH and small SDHs, but headaches were the predominant symptom in the overwhelming majority.[6] As our patient had only radiological findings suggestive of SIH, the positive outcome was assessed by SDH regression. Likewise, Chaudhary et al. reported a case of recurring SDHs despite surgical evacuations, and in whom a potential leak was found in the thoracic spine, with possible benefit to subsequent EBPs.[7]

Interestingly, our patient's response to topiramate, as opposed to levetiracetam, supports the increasingly recognised efficacy of this agent on cortical spreading depression, which is the presumed pathophysiological mechanism of paroxysmal neurological events in patients with SDHs.[8] It is also worth noting that topiramate's ability to decrease CSF production, through the inhibition of carbonate anhydrase, did not in our case interfere with the positive outcome.

A limitation, in this case, is that the CSF leak was not proven. We elected not to perform CT or radioisotope myelography as it was felt that these techniques could, by themselves, cause a CSF leak. Empirically, our plan was to consider myelography if the initial EBP was ineffective in order to perform a more localised one afterwards. CSF may have leaked from one of the several cysts, but it is also possible that these merely represent dural friability, which predisposed the patient to a small remote leak. Another hypothesis is that the patient's SIH was not due to a CSF leak, but due to CSF pooling. Nevertheless, EBP was thought to create intrathecal volume replacement, which could have been sufficient to prevent further SDH deterioration. The latent effect

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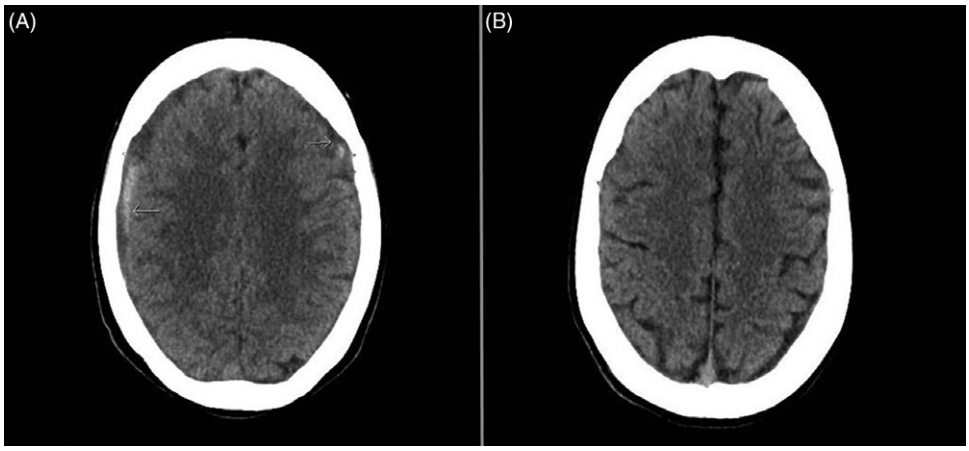

Figure 1: *(A) Initial bilateral SDHs on head CT showing acute (arrows) on chronic collections. They measured maximally 6 mm on the right and 3 mm on the left. (B) Shows resolution of the collections 2 months after epidural blood patch.*

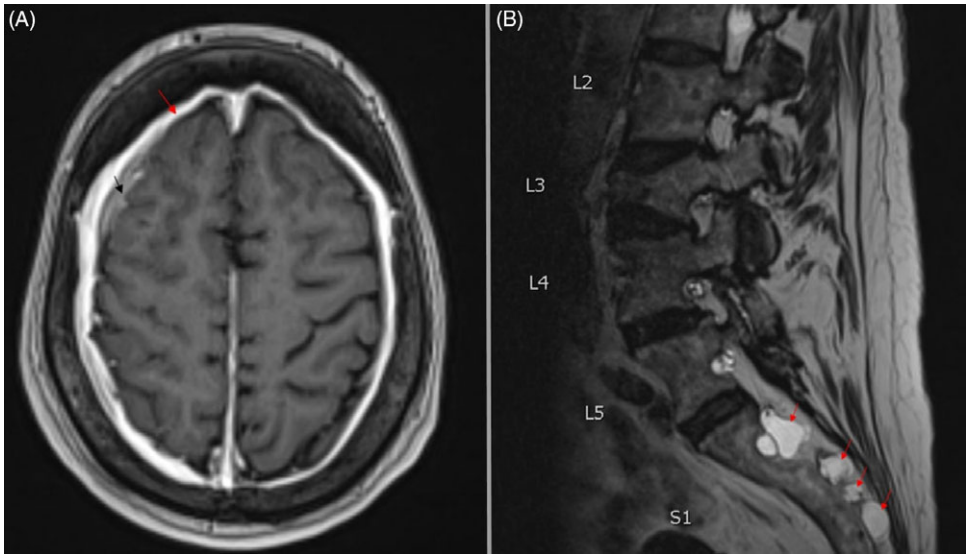

Figure 2: *(A) Axial T1 brain MRI with contrast shows pachymeningeal enhancement (red arrows) and subacute SDHs (black arrows), measuring up to 10 mm on the right and 4 mm on the left. (B) T2 sagittal lumbar and sacral MRI shows multiple meningeal cysts (arrows).*

resulting from sealing the leak may or may not have been present as a leak has never been proven. However, as there has been no SDH reaccumulation on follow-up, a successful seal is likely.

In conclusion, our case suggests that SIH should be suspected in cases of atraumatic SDH, even in the absence of orthostatic headaches. Acephalgic SIH has only been anecdotally been reported, but it remains possible that such an entity may be underreported due to a lack of medical awareness and underdiagnosis. Radiological findings such as smooth dural enhancement may be sought on MRI to support the diagnosis. In such cases, clinicians should look for a CSF leak and it may be reasonable to consider the first EBP before proceeding to more invasive investigations and treatments. Ultimately, our case highlights a benefit of EBP on an acephalgic SIH by effectively resolving SDHs and their underlying neurological spells.

Acknowledgements

All authors declare that this work is the product of an original case that was encountered at their facility. The sources used are those listed in the bibliography and identified as references.

Disclosures

The authors have no disclosures.

Statement of Authorship

MRB has done the required academic literature review for the conception of the article and drafted the article. PB has done a critical revision of the article and performed the final approval of the version to be published.

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