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Case Report

Acute subdural hematoma recurrence during drain removal associated with spontaneous intracranial hypotension – A non-reported complication

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ABSTRACT

Background: Spontaneous intracranial hypotension (SIH) is an uncommon, benign, and generally self-limiting condition caused by low cerebrospinal fluid (CSF) volume and pressure usually caused by a CSF leak. Patients with SIH have an increased incidence of subdural hematomas (SDH), which may be bilateral and recurrent.

Case Description: We report a unique case of a man presenting with SIH and bilateral SDH that were drained with bilateral craniotomies. During drain removal, the patient had an acute neurological deterioration and a CT scan showed SDH recurrence. The patient had two new recurrent SDH afterwards. After the third surgical intervention, the drain was removed in the OR with concomitant subdural saline infusion, there was no recurrence of SDH after that and the patient has had no further complications after a 2-year follow-up.

Conclusion: Patients with intracranial hypotension are predisposed to form SDH. In this case, drain removal caused further decrease in intracranial pressure and triggered a new SDH formation, subdural saline irrigation masked atmospheric pressure and prevented this complication from happening again.

Keywords: Drain removal, Spontaneous intracranial hypotension, Subdural hematomas, Subdural irrigation

INTRODUCTION

Spontaneous intracranial hypotension (SIH) is an uncommon, benign, and generally self-limiting condition caused by low cerebrospinal fluid (CSF) volume and pressure usually caused by a CSF leak.[9] This process results in a downward traction of the brain, that may cause headaches, subdural fluid collections, and possible brain herniation, among other complications.[2,10] Approximately 16–57% of patients with SIH are expected to suffer from subdural hematomas (SDH), and it is particularly predominant in males. Case series have reported mixed clinical outcomes that span from a benign and self-limiting course to death from SDH. There is an increased rate of SDH recurrence with intracranial hypotension, and SIH treatment prior or after SDH management still remains controversial.[12] We report a unique case of a man presenting with SIH and bilateral SDH that recurred twice spontaneously after subdural drain removal; after the third surgical intervention, the drain was removed in the OR with subdural saline infusion without further complications.

CASE REPORT

History of present illness and first intervention

A 48-year-old man presented with an orthostatic headache of 3 months duration that progressively worsened for the past few hours, along with somnolence and vomiting. There was no significant medical history. A noncontrast head CT scan was ordered, showing bilateral subacute SDH [Figure 1a]. He was admitted and a bilateral craniotomy for hematoma drainage and drain placement was successfully performed. The patient's evolution was uneventful with a normal postoperative physical examination and control CT scan [Figure 1b].

Drain removal and second intervention

The drain on the right side was removed 24 h after surgery at a bedside procedure; moments afterwards the patient suddenly presented with a headache, vomiting, tonic posture, right ocular deviation, and an altered mental status. A new CT scan showed recurrence of the SDH on the left cerebral hemisphere, contralateral to the drain that was removed [Figure 1c]. The patient was taken to the OR where the hematoma was drained again. The hallmarks of the postoperative evaluation were a mild headache and improvement of neurologic signs and symptoms. The drain was removed without any complications.

Diagnosis of SIH

Based on characteristic clinical findings of orthostatic headaches and SDH, a differential diagnosis of SIH was established, and an MRI was performed. The diagnosis was confirmed with classic findings of diffuse pachymeningeal enhancement, enlargement of pituitary gland, engorgement of sigmoid sinus, brainstem sagging and effacement of the suprasellar and prepontine cisterns, as well as a decreased mamillopontine distance. However, a CT myelography did not show findings suggestive of CSF leakage.

Third intervention and remission

Three weeks after the second surgical intervention, the patient developed somnolence, hiccups, positional headache, oculomotor nerve palsy, and respiratory failure requiring assisted mechanical ventilation. An epidural blood patch (EBP) procedure targeting thoracolumbar spinal cord was done, with no change in the clinical outcome. A new CT scan showed recurrence of the left SDH and a new intervention for hematoma drainage with drain placement was scheduled [Figure 1d]. Postoperative evolution was again uneventful with good clinical outcome and improvement of neurologic signs and symptoms. During drain removal, it was decided that the patient be taken to the operative room, where normal sterile saline was infused into the subdural space at the same time the drain was being removed. The patient was discharged with a normal CT scan [Figure 1e]. For follow-up, the patient had an MRI scan after 5 months, with no findings related to intracranial hypotension or SDH [Figure 2] and there was no recurrence of neurologic signs after 24 months.

DISCUSSION

SIH is caused by a CSF leak that gives rise to intracranial hypotension; its main clinical manifestation is an orthostatic headache that can be accompanied by other symptoms including nausea, vomiting, neck stiffness, or ocular nerve palsies.[14] The diagnosis is made with the measurement of CSF pressure and classic imaging signs on the MRI or CT myelogram. A conservative treatment is usually enough to manage the remission of SIH, if this does not work an EBP can be used.[13] However, patients may develop bilateral subdural collections that require surgical drainage due to acute clinical deterioration. Loss of CSF results in the formation of compensatory subdural fluid collections which may enlarge the subdural space and tear bridging veins, resulting in a SDH formation.[7,15] The main clinical sign of these patients is an impaired level of consciousness that spans from somnolence to coma; it can be attributed to either

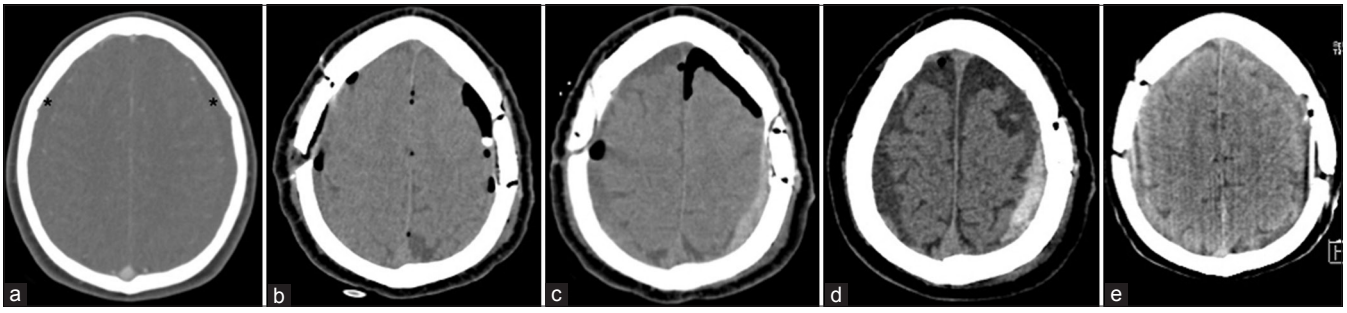

Figure 1: CT scans showing the patient's evolution. (a) Initial CT scan showing bilateral subdural hematomas. *(b) Postoperative CT scan showing bilateral mini-craniotomy with associated pneumoencephalus. (c) Acute left subdural hematoma after the right drain removal. (d) Postoperative CT scan taken after the first recurrence and second surgery, showing the left subdural hematoma that appeared 3 weeks after the second drain removal. (e) Final CT scan, the patient had no further subdural hematoma recurrences.

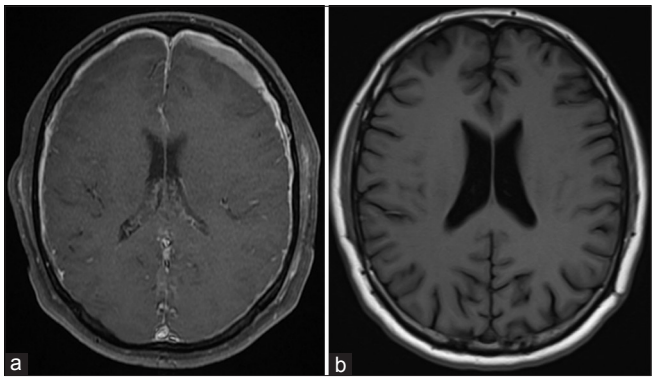

Figure 2: Preoperative and postoperative MRI scans. (a) Gadolinium-enhanced coronal head MRI showing a left subdural hematoma and diffuse pachymeningeal enhancement, taken after the first surgery and drain removal. (b) Control MRI taken 5 months after the third surgery, there is no recurrence of subdural hematoma or signs of intracranial hypotension.

intracranial hypotension (brain sagging) or the compressive effect of the hematoma.[3,11] It is important to differentiate patients with "primary" SDH and SDHs due to SIH. Patients with SIH will most likely be younger (median age of 40) and have additional findings on CT scans such as brain sagging and pseudo-subarachnoid hemorrhage features.[8]

Acute clinical manifestations triggered by the drain removal in our case show the dual presentation of SIH. The patient presented with somnolence, vomiting, and a severe headache that remitted with hematoma drainage, suggesting a compressive origin of his manifestations. It is hypothesized that drain removal causes a direct communication between the atmosphere and intrathecal space, and the atmosphere pressure further decreases the intracranial pressure in predisposed patients with SIH.[6] The previous scenario would cause either clinical manifestations from brain herniation or recurrence of SDH with acute signs and symptoms. Due to the clinical improvement of our patient after the second hematoma drainage, the drain removal most likely caused a decrease of intracranial pressure with symptomatic hematoma recurrence.

After the third hematoma drainage, we did a thorough analysis to implement a technique that allowed drain removal without subsequent intracranial hypotension. To avoid further decrease of the intracranial pressure and mask the effect of atmospheric pressure, we infused saline solution into the subdural space simultaneous to drain retraction. The patient did not course with worsening signs and symptoms and has been asymptomatic for 2 years.

It is important to acknowledge that bridging veins can be teared as the drain is removed and subsequently caused a hemorrhage into the subdural space. Although it is a possibility, Alcala-Cerra *et al*. reported no difference in hematoma recurrence between patients with chronic SDH treated with and without subdural drain after hematoma drainage in seven randomized clinical trials, further raising the question of the relevance of the relationship with bridging veins – drain. SDH recurrence in patients with SIH could represent many etiologies, and the propensity of patients to develop this condition could mean a low resistance threshold for all the possible causes of SDH.[1]

García-Morales *et al*. reported a case with four surgical interventions for symptomatic recurrent SDH in a patient with SIH; although no relationship was specified between hematoma recurrences and drain removal, conservative treatment for SIH and subsequent hematoma drainages was the hallmarks of the case.[5] Having said this, and analyzing the similar workup of our case, it is important to mention the significance of the EBP procedure in our patient after SIH diagnosis, for no clinical improvement was noted at first. The importance and clinical relevance of subdural saline irrigation should be further evaluated since definite hematoma remission could have been accomplished due to the EBP/SDH drainage combined approach, not the irrigation method *per se*. In a case series of 23 patients with SIH and SDH, Takahashi *et al*. concluded that symptomatic SDH should be treated with surgical intervention immediately after EBP procedure.[12] The same recommendation is done by Ferrante *et al*. in a report of 35 cases, with the addition of surgery before EBP if the hematoma evacuation was emergent; in this case, EBP should be done before the patient gets up.[4]

CONCLUSION

SIH is a frequently misdiagnosed disorder with a myriad of neurologic signs and symptoms. Patients are prone to develop recurrent SDHs that are difficult to treat. No standardized treatment approach has been established for this type of presentation as some centers perform EBP either before or after hematoma evacuation, if the latter is required. We present a novel approach for the treatment of recurrent SDH in patients with SIH, clinical resolution was achieved with transoperative subdural saline infusion, as the effects of the direct communication of atmospheric pressure and intracranial cavity can be lessened by fluid volume compensation. Further, consensus needs to be reached as the prognosis of these patients can be largely impacted by an approach proven to be safe and free of long-term complications.

Ethics approval

This report was approved by Comite de Bioetica y Cuidados Paliativos HCG-ING Ethics Committee.

Reference number: 2020/0301.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

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