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Case Report

When does too little pressure become too much? A case of spontaneous intracranial hypotension presenting with acute loss of consciousness

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ABSTRACT

Background: We present a unique case of spontaneous intracranial hypotension (SIH) presenting with acute collapse and loss of consciousness.

Case Description: The affected patient suffered an abrupt decline in level of consciousness several weeks after initial diagnosis. The patient was urgently transferred to a specialist neurosurgical unit. Imaging showed bilateral subdural fluid collections with significant associated local mass effect. The treating team faced a clinical conundrum with a lack of clarity as to whether this sudden deterioration was secondary to the local pressure effect on brainstem traction from reduced intracranial pressure. A decision was made to proceed with urgent burr-hole decompression of the bilateral subdural fluid collections.

Conclusion: After a protracted, complex postoperative course, the patient recovered to full functional independence. To the author's knowledge, this is the first case in literature describing successful surgical management of SIH, with bilateral burr-hole evacuation to relieve the paradoxical mass effect of bilateral subdural fluid collections.

Keywords: Cerebrospinal fluid effusion, Essential intracranial hypotension, Neurosurgery, Spontaneous subdural surgical management

INTRODUCTION

Spontaneous intracranial hypotension (SIH) is characterized by postural headache, often secondary to an underlying idiopathic spinal cerebrospinal fluid (CSF) leak, resulting in reduced intracranial pressure. It has an estimated annual incidence of 5/100,000 of the population.[10] This figure is likely under-representative, with SIH being under-diagnosed, due to its subtle symptoms and benign course.[9]

Management of SIH is usually conservative, with hydration and bed rest. Further, intervention by means of an epidural blood patch may be pursued following failed conservative management. Surgical intervention is rarely performed. Repair of dural defects or CSF venous fistulae has been described but is exceedingly rare.[3]

We report a case of SIH presenting with acute collapse and loss of consciousness. There are no published reports of SIH presenting with acute deterioration and coma managed with urgent decompressive surgery. This case presents a unique diagnostic dilemma, with uncertainty as to whether a drop in Glasgow Coma Score (GCS) was due to brainstem herniation through the foramen magnum secondary to low intracranial pressure or a result of the mass effect of subdural fluid collections on the cerebrum. A decision was made to urgently evacuate the fluid collections. The patient made a complete recovery following the intervention.

CASE HISTORY

A 48-year-old right-hand dominant male presented to a local emergency department with a 6-week history of headaches and ataxia which began after weightlifting. Headaches were described as intermittent and worse with standing. Background history was unremarkable with no comorbidities, prior surgery, or recent trauma. Following admission, routine hematological tests were unremarkable. Work-up also included computed tomography (CT) brain and magnetic resonance imaging (MRI) of the brain with contrast. Imaging revealed bilateral hypoattenuating subdural collections with associated bifrontal sulcal effacement on CT [Figure 1]. MRI confirmed these to be fluid collections, with diffuse meningeal enhancement on T1-weighted sequences suggesting intracranial hypotension as the underlying cause [Figures 2a and b]. An MRI spine demonstrated epidural fluid collections in the cervicothoracic region [Figure 3]. A diagnosis of SIH was made, reinforced by classical history and imaging findings. The patient was managed conservatively and discharged.

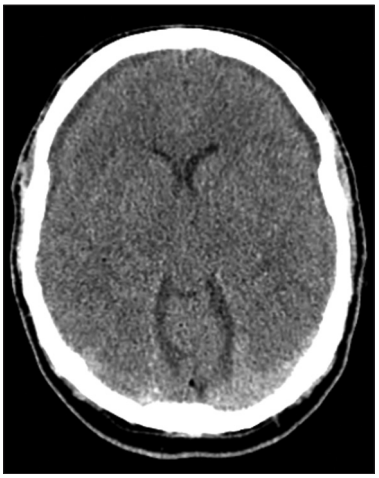

Figure 1: Bifrontal hypoattenuating subdural collections on axial noncontrast computed tomography (CT) brain.

Several weeks later, the patient was found collapsed at home. He was difficult to rouse and was brought by ambulance to a local hospital. A GCS of 14 was recorded on arrival. This subsequently dropped to 10. An urgent CT brain exhibited enlarging subdural fluid collections, particularly on the right [Figure 4], with an increased local mass effect. The neurosurgical service at our tertiary referral center was consulted and the patient was immediately transferred. A decision was made for the urgent burr-hole evacuation of the subdural fluid collections. Bilateral frontal burr holes under general anesthetic released these collections under high pressure and the patient was transferred to the intensive treatment unit.

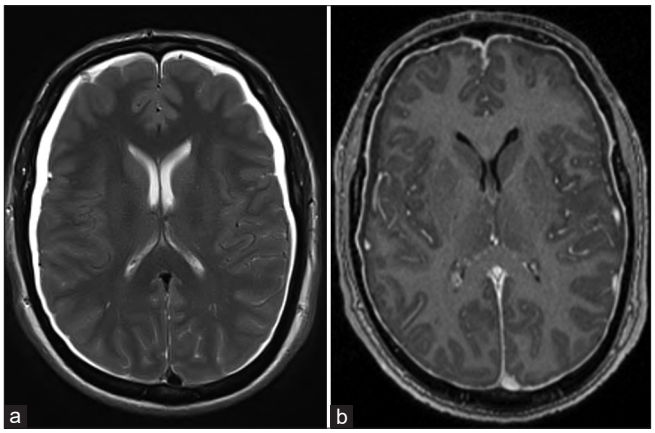

Figure 2: (a) High-signal fluid collections on axial T2-weighted magnetic resonance imaging (MRI) brain. (b) Diffuse pachymeningeal enhancement on axial T1 post contrast MRI brain, indicative of intracranial hypotension.

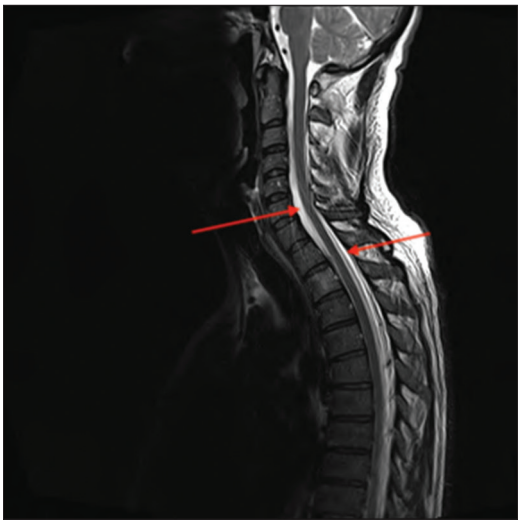

Figure 3: Sagittal T2-weighted magnetic resonance spine demonstrating anterior and posterior epidural fluid collections (red arrows) in the cervicothoracic region.

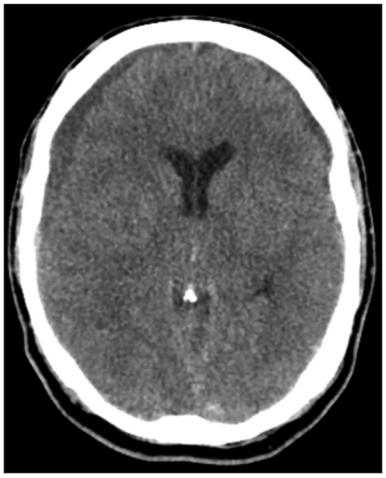

Figure 4: Repeat computed tomography (CT) brain with increasing size of right-sided subdural collection.

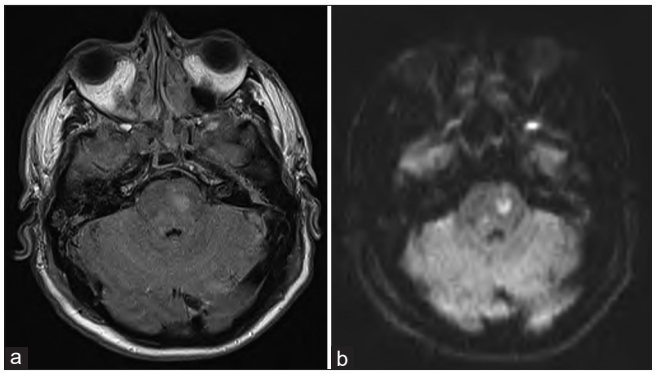

Figure 5: Magnetic resonance imaging brain (MRI) demonstrating (a) focal area of hyperintensity in the left pons on axial fluid attenuated inversion recovery and (b) corresponding diffusion restriction on diffusion-weighted imaging.

A complex postoperative course followed. The neurosurgical team advocated for the patient to be nursed in the Trendelenburg position. However, the patient was often managed in a head up position for the management of respiratory problems. A blood patch performed to seal a hypothesized leak at the cervicothoracic spine yielded no clinical improvement 4 days after surgery. Spinal epidural fluid collections remained stable on MRI. GCS remained low; however, the patient began flexing in response to pain with severe right-sided weakness. An up-to-date MRI brain [Figure 5] demonstrated a diffusion-restricting lesion in the left pons, with features concerning for an acute brainstem infarct.

Despite the grim prognosis, a repeat blood patch was performed 2 weeks after the first. The patient was strictly nursed in a continuous Trendelenburg position following this. This yielded encouraging results, and GCS rose to 14 with successful extubation 2 days later. The cervicothoracic epidural fluid collections gradually reduced in volume over serial MRI [Figures 6a-c]. GCS improved to 15 and the patient was discharged 1 week after returning to the ward.

At follow-up 2 months post discharge, the patient was fully independent with only a partial right trochlear nerve palsy and mild right-sided paresthesias, having regained full power on both sides.

DISCUSSION

SIH is uncommonly encountered. It has an estimated annual incidence of 5/100,000.[10] Etiology and pathophysiology are poorly described, due to their rarity. No clear risk factors or predisposing conditions have been defined in the literature. Patients classically present with orthostatic headache, although atypical presentations have

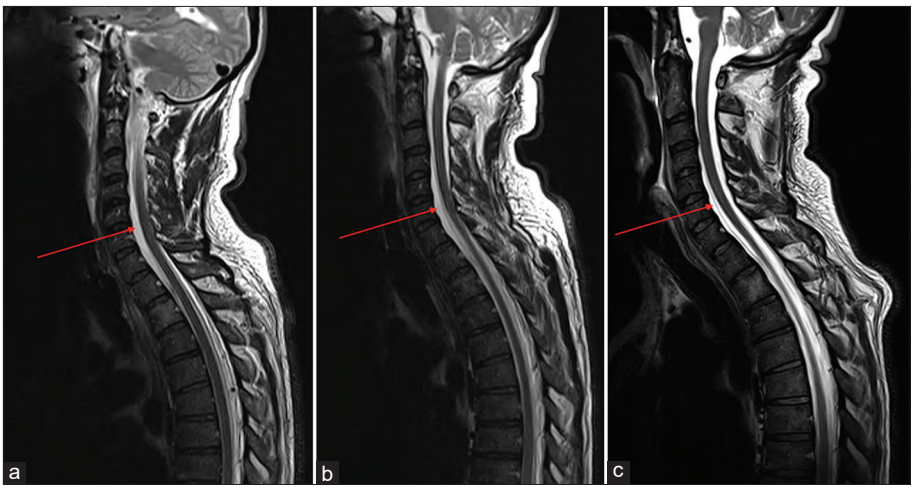

Figure 6: Serial magnetic resonance imaging spines demonstrating steadily reducing epidural fluid collections (red arrows) on (a) day 1 postoperative (b) day 1 post first blood patch, and (c) day 1 post repeat blood patch.

been described, which include neurology that does not correlate with image findings, Parkinsonian symptoms, and dementia with frontotemporal features.[1] The presentation can vary depending on patient demographics, underlying medical conditions, and the chronicity of subdural collections. Awareness of this helps to enable the recognition of atypical cases of SIH. Diagnosis is often clinical and may be reinforced with findings on MRI, with diffuse pachymeningeal enhancement and subdural fluid collections the most common findings.[6] Physical examination is also important. Associated focal neurology is not uncommon. Sixth cranial nerve palsy constitutes the most frequent finding and is thought to be secondary to traction on the cisternal portion of the abducens nerve due to brain sagging. Nonspecific visual field defects have been reported. Fundoscopy should be performed to assess for paradoxical papilloedema, which has also been described.[5] Imaging is particularly important in establishing diagnosis in atypical cases.[1] The prognosis is generally excellent. The condition usually follows a benign course, with conservative management proved successful in almost all cases.

Our case has a distinct course. Acute collapse confounded the clinical picture and presented a unique challenge in deciding the most appropriate management. While the patient had known SIH, it was impossible to determine whether the rapid deterioration was secondary to brainstem herniation through the foramen magnum from sagging related to low intracranial pressure or due to the mass effect of subdural fluid collections on the cerebrum and subsequent coning. Given the sudden acute decline and considerable increase in the size of the bifrontal subdural fluid collections, a decision for urgent bilateral burr-hole decompression was made. While several cases of SIH presenting with coma have been described,[8] there has been little published to describe the use of surgical decompression in this setting, with only three previous successful cases to the author's knowledge.[2,7] The rapid clinical deterioration, in this case, mirrors the course of previously published cases of other spontaneous nontraumatic subdural collections, successfully managed with urgent decompression.[4] This is the first report of successful management with bilateral burr-hole decompression to relieve the paradoxical mass effect. There is also a paucity of literature describing the management of patients with refractory SIH in the Trendelenburg position. Our case highlights the importance for this to be considered in complex scenarios.

CONCLUSION

After a long, protracted and complex postoperative course, the patient recovered to full functional independence. To the author's knowledge, this is the first case in literature describing successful surgical management of SIH, with bilateral burr-hole evacuation to relieve the paradoxical mass effect of bilateral subdural fluid collections.

Declaration of patient consent

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

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Nil.

Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The author(s) confirms that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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