Spontaneous intracranial hypotension (SIH) is a relatively rare entity with an estimated incidence of 5 in 100,000 and it is uncommonly encountered in neurosurgical practice. The classic triad of SIH includes orthostatic headache, diffuse pachymeningeal enhancement on magnetic resonance imaging (MRI) with gadolinium, and low cerebrospinal fluid (CSF) pressure. A handful of reports have described delay in diagnosis or misdiagnosis of SIH due to varied clinical presentations. Although the clinical course in SIH is deemed to be benign, complications are not infrequent. SIH may be associated with the descent of the brainstem, subdural hygroma, or hematoma. Surgical drainage for subdural hematoma has been recommended in acute clinical deterioration but few cases reported acute neurological deterioration after craniotomy in the setting of SIH. The delay in diagnosis of SIH and surgical drainage for subdural hematoma might lead to disastrous complications, as encountered in the present case.

Case Report

A 36-year-old male with intermittent history of headache since 4 weeks presented with acute onset excruciating headache to our emergency department. The headache was generalized, continuous, squeezing with no relief, or aggravation on lying down. Past history was unremarkable, and there was no history of head trauma. On examination he was alert with no neurological deficit. No nuchal rigidity was noted. Blood examination revealed a normal hemoglobin and coagulation profile. Computed tomography (CT) scan findings of bilateral chronic subdural hematoma (SDH) collection with increased attenuation along basal cistern raised the possible diagnosis of acute subarachnoid hemorrhage (SAH). Intracranial angiography revealed no evidence of aneurysm or vascular malformations. Conservative treatment including bed rest and careful observation was done. Until then, the diagnosis of SIH was not made, and follow-up intracranial angiography was planned after a week considering the possibility of concealed aneurysm. In subsequent days, the patient’s consciousness deteriorated progressively to be lethargic which was interpreted as a sign of increased intracranial pressure due to bilateral chronic SDH. After confirming no aneurysm on follow-up...
CT angiogram, the patient underwent emergency bilateral burr hole drainage of hematoma. The intraoperative course was unremarkable. Immediate postoperative CT scan demonstrated poor visualization of posterior fossa cisterns [Figure 2a, left] with large pneumocephaly in both cerebral hemispheres [Figure 2a, right]. After the recovery period, the patient did not regain consciousness; instead, there was progressive worsening of consciousness. The 2-h follow-up CT scan revealed a huge epidural hematoma on the left side [Figure 2b]. An emergency craniotomy and hematoma evacuation was performed. After the second surgery, the patient was deeply stuporous without any clinical improvement. The 2-day follow-up diffusion weighed magnetic resonance imaging (MRI) evidenced acute cerebral infarction on bilateral territories of posterior cerebral artery and CT scan showed poor visualization of the fourth ventricle [Figure 2c, left] with supratentorial pneumocephalus and subdural fluid collection [Figure 2c, right]. Unusual rapid clinical deterioration despite prompt hematoma evacuation, poor visualization of the fourth ventricle, and pneumocephalus on imaging were now interpreted as the result of downward displacement of the brain, and an underlying spinal CSF leak was suspected. The patient was placed in the Trendelenburg position and an empirical epidural blood patch was applied using a 10 ml autologous blood, one each for a cervico-thoracic and thoraco-lumbar junction. A rapid improvement in consciousness (from Glasgow coma scale score 7-11) was noted over the period of 3 days after the procedure and the CT scan showed well visualization of the fourth ventricle [Figure 2d]. The patient demonstrated full clinical recovery, 4 weeks after the epidural blood patch. A complete spine CT myelography performed to evaluate the possibility of remaining CSF leak demonstrated small leaks at cervical C4-5 level [Figure 3a]. Because the patient was alert and had no symptoms associated with SIH, no further intervention was performed. The patient was discharged and the 4-month post-operative follow-up brain MRI showed no evidence of subdural fluid collection [Figure 3b] or brain herniation. Bilateral cortical blindness due to bilateral occipital infarction and mild cognitive deficit gradually improved 8 months after the surgery.
Discussion

SIH is a benign entity that is rarely encountered in the neurosurgical practice. The clinical features of orthostatic headache and the specific MR findings of diffuse enhancement of the pachymeninges, venous engorgement, pituitary enlargement, brain stem herniation, and subdural collection are clues to diagnose SIH. Nevertheless, the first imaging study performed when patients with acutely aggravated headache arrive in an emergency room is usually a CT scan. The initial CT scan findings of increased attenuation in basal cisterns along tentorium cerebelli resemble SAH, addressed as pseudo-SAH. The incidence of pseudo-SAH in patients with SIH is reported to be 10%, making it a less common finding in SIH.[13]

SIH usually resolves spontaneously by conservative treatment such as strict bed rest and hydration therapy.[1] The epidural blood patch and Trendelenburg position are treatment of choice when the symptoms are persistent and refractory.[1,2,8] Subdural fluid collections are common in patients with SIH, varying in appearance from thin subdural hygromas to large SDH.[2,6,13] The reported incidence of SDH and non-hemorrhagic subdural fluid collections in association with SIH is from 10% to 69%.[2,6,13] Loss of CSF results in the formation of compensatory subdural fluid collection, which might gradually enlarge the subdural space resulting in tearing of bridging veins.[6] The development of SDH in patients with SIH has been associated with constant and severe headache, acute deterioration of symptoms, associated with mass effect and midline shift resulting in decreased consciousness.[9] The treatment modality for SDH in SIH has no general consensus. Subdural hematoma or hygroma, which is of limited volume, will usually resolve spontaneously after the normal CSF volume has been restored.[1,4] The associated chronic SDH can deteriorate with time necessitating neurosurgical intervention.[1,5] Some reports have recommended evacuation of hematoma presenting with progressive enlargement of SDH.[6,8,9] Other reports have suggested the possibility that the evacuation of SDH in patients with low CSF volume can worsen the downward shift of the brain with buckling of brain stem and coma[10,11] or multiple recurrences.[14] Craniotomy may decrease the buoyant effect of CSF in keeping the brain afloat more; consequently, a position-dependent caudal herniation of the brain might occur in SIH.[6]

In the present case, the atypical clinical presentation and imaging signs led to delayed diagnosis of SIH. We believe that preoperative finding of pseudo-SAH is due to brain sagging, as previously reported,[13] which was aggravated to downward brain herniation after burr hole drainage and craniotomy. Clues suggesting SIH rather than true SAH or SDH in this case would be absent nuchal rigidity, lower attenuation values on CT scan within the basal cisterns compared to the true SAH, and large pneumocephaly, considering his age, after the burr hole drainage.

Conclusion

The present case shows that the possibility of SIH should be considered when evaluating the patient with SAH with bilateral chronic SDH on initial CT scan. Brain MRI with gadolinium enhancement or lumbar tapping will render critical clues for early diagnosis of SIH, which might obviate unnecessary multiple imaging and surgical intervention. In addition, it should be emphasized that surgical drainage of the subdural hematoma in SIH, before correcting CSF leaks, might result in serious complications.

References

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