CASE REPORT DOI: 10.15713/ins.bhj.154

Spontaneous Intracranial Hypotension with Chronic Subdural Hematoma-A Case Report of Multimodality Management

ABSTRACT

Aim: We would like to present our experience in managing spontaneous intracranial hypotension (SIH) with chronic subdural hematoma (SDH), which required multimodality treatment. **Background:** SIH usually occurs due to a cerebrospinal fluid (CSF) leak, causing an overall decrease in CSF volume. The incidence reported is 5/100000 population per year but lacks typical symptomatology. It can occur in any age group with a female preponderance ratio of 1.5-2:1. CSF leaks may be due to dural tears, meningeal diverticulum, and CSF-venous fistulas in the dorsal-lumbar region. SIH causes sagging of the brain, causing subdural spacing and SDH. **Case Description:** A middle-aged male patient presented with an orthostatic headache with a history of epidural catheter use in the past. After a detailed clinical evaluation, the patient was advised for an MRI brain with whole spine screening, which showed chronic SDH with thoracic epidural CSF collection. The patient was evaluated and underwent multimodality treatment with surgical burr hole evacuation of SDH, an epidural blood patch for epidural CSF leak, and medical management with Tranexemic Acid. On the 4-week follow-up, patient did not have any symptoms, and the 9-month follow-up showed no symptoms and no recurrence on the CT scan. **Conclusion:** SIH with SDH usually needs multiple modes of treatment. Our step-up plan from conservative to multimodality management benefitted the patient with a complete resolution of symptoms and a complete resolution of SDH.

Key words: SIH with SDH, Cerebrospinal fluid (CSF), Epidural blood patch (EBP)

INTRODUCTION

Spontaneous intracranial hypotension (SIH) occurs due to a spontaneous cerebrospinal fluid (CSF) leak from the spine, causing a decreased CSF volume and an orthostatic headache. Decreased CSF volume causes sagging of the brain and increased subdural space. As per the Monre-Kelle doctrine, if CSF decreases, other two components may increase namely the brain and blood. Since the brain cannot increase in volume and compensate for CSF volume, blood replaces it, causing subdural hematoma (SDH).

SIH has an incidence of 5/100000/year and can occur at any age. It has a female preponderance with a ratio of 1.5-2:1.[1] Connective tissue disorders like Marfan's syndrome or Ehler–Danlos syndromes have higher risks of SIH.[4]

Spinal CSF leaks may occur daily due to three mechanisms.[1]

1. Type 1 dural tear (maybe ventral or dorsal): In spontaneous dural tear, there may be a small bony spicule causing dural tear, mostly ventrally. There can also be an iatrogenic cause due to a lumbar puncture or epidural catheters, mostly dorsally.[1]
2. Meningeal diverticulum (which may be simple or complex): Due to rupture of herniated arachnoid through focal dural weakness in the axillary sleeve region of the nerve root, causing CSF leak. Sometimes there may be slow and chronic seeping of CSF from this, which causes SIH.[1]
3. CSF-Venous fistula: abnormal fistulous communication between the perineurial subarachnoid space and surrounding veins causes direct CSF leakage in the venous system, causing SIH. The Most common location is between T7-T12, and 82% are also associated with meningeal diverticulum.[1]

Apart from three types mentioned above, there may be an indeterminate or unknown type.[1]

Schievink's diagnostic criteria are as follows:[1]

a. Demonstration of spinal CSF leak (presence of extrathecal CSF). If A criteria is not met:
b. Cranial MRI changes of intracranial hypotension (SD collection, dural enhancement, and sagging of the brain.) with atlas one of the below-mentioned criteria

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1. Low opening pressure (<60 mm H_2O)
2. Spinal meningeal diverticulum
3. Improvement of symptoms after epidural blood patch (EBP)

If both criteria A and B are not met, then C

c. Presence of all or at least two of the following if the typical orthostatic headache is present.
   1. Low opening pressure (<60 mm H_2O)
   2. Spinal meningeal diverticulum
   3. Improvement of symptoms after EBP

Symptomatically, the patient initially may have a classical orthostatic headache; the patient may also have an unexplained chronic headache, which needs to be evaluated and investigated further. Around 94% of SIH is usually misdiagnosed as migraine or meningitis, as per Dobrocky *et al*. Hence, these types of non-specific chronic headaches should also be evaluated for SIH.

Due to a decrease in CSF volume, which causes sagging of the brain and subdural collection, or SDH.[6] SDH may be small or big enough to cause mass effects and neurological deficits. Hence, treatment for SIH and SDH can be simple conservative management, EBP, burr hole, craniotomy, or evacuation of SDH.[6] For conservative management of SDH, many drugs have been used; one such drug is tranexemic acid.[3] It daily decreases plasmin activity by reversibly binding to lysine sites on plasminogen and reducing both inflammation and fibrinolysis.[3] Medical management can be done in symptomatic or mildly affected SDH patients; however, if there is a mass effect or significant neurological deficits, then burr hole evacuation, craniotomy, and clot/collection evacuation can be done. Another important component of the management of SIH is strict flat in bed rest post-blood patch alone or with SDH evacuation.

CASE REPORT

A 53-year-old male was operated on for right renal cell carcinoma and has presented with headaches for 10 days. The patient had headaches mainly in the sitting or standing position and was relieved in the lying supine position. Associated with multiple episodes of vomiting. The patient had an epidural catheter for preoperative analgesia. There is no other significant history.

On examination, the patient was conscious and oriented. Normal eye movements. Pupils bilaterally were 2 mm and reactive to light. Deep tendon reflexes were normal (+2) with a negative Banbinski. There was no pronator drift, and other neurological examinations were within the normal limit.

The patient underwent radiological investigations [Figures 1-3].

With detailed clinical and radiological evidence of SIH, chronic SDH, and epidural CSF collection in the D9-D12 epidural space, the patient underwent an EBP. Following the procedure, the patient was advised to remain in a completely flat bed rest position. However, after 72 h, as the patient gradually transitioned to a head-up position day by day, they started experiencing a similar type of headache.

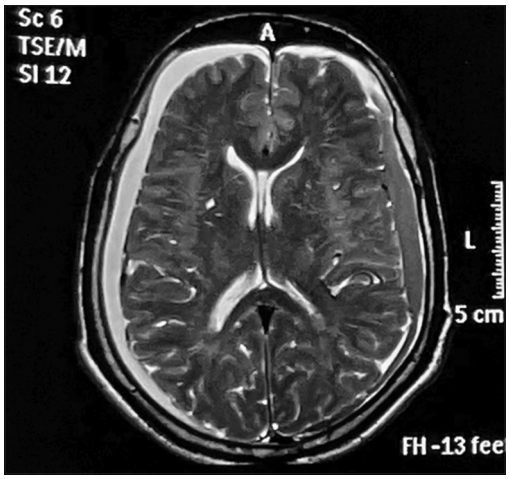

Figure 1: MRI brain T2 showing bilateral Subdural hematoma

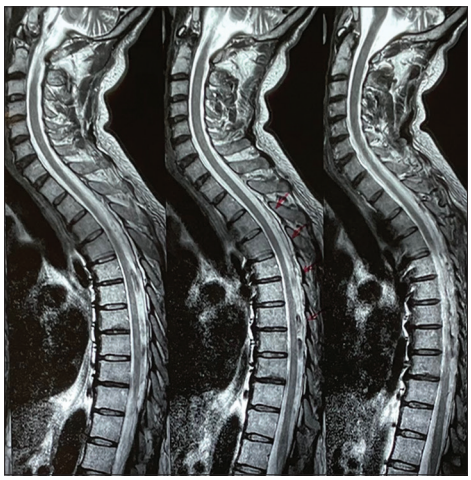

Figure 2: T2 saggital view showing dorsal epidural CSF collection

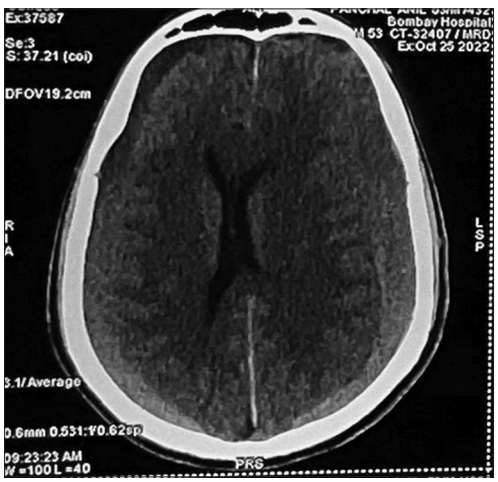

Figure 3: CT brain showing bilateral SDH

Subsequently, the patient underwent another EBP, and an extended period of complete bed rest (1 week) was recommended. A CT scan of the brain was performed to assess any changes in the size of the SDH, revealing a significant increase. Hence, burr hole evacuation of the SDH was performed, resulting in significant improvement, and the orthostatic headache was resolved. The patient was once again advised to rest and was discharged.

However, after 1 week, the patient presented with a recurrence of orthostatic headache, accompanied by left-sided weakness. Upon examination, the patient exhibited a left pronator drift, and a CT scan of the brain showed SDH with compression. A combined approach, involving the evacuation of the SDH and repair of the spinal CSF leak, was planned. To localize the site of the dural tear, an MRI of the spine was performed. However, the leak's exact location could not be identified but was localized to the lower dorsal region, leading to a targeted EBP along with subdural collection evacuation.

Following which, the patient was started on Tranexamic Acid and advised to complete the flat bed rest position for 7 days. During this period, the patient experienced no orthostatic headache, and the weakness also resolved. Consequently, the patient was discharged and instructed to adhere strictly to bed rest and avoid strenuous activities. One month later, the patient returned for a follow-up examination, reporting no headache or limb weakness, and a 9-month follow-up CT scan showed complete resolution of the SDH (Figure 4).

DISCUSSION

The Schievnik criteria are valuable in diagnosing SIH, as they have been elaborated upon.[1] Understanding the classification of spontaneous spinal CSF leaks is essential for managing SIH, as it helps determine the anatomical pathology of the spinal CSF leak.[1] Type 1 involves a dural tear, which can be ventral (1a) or dorsal (1b), with ventral tears being more common in the thoracic spine.[1]

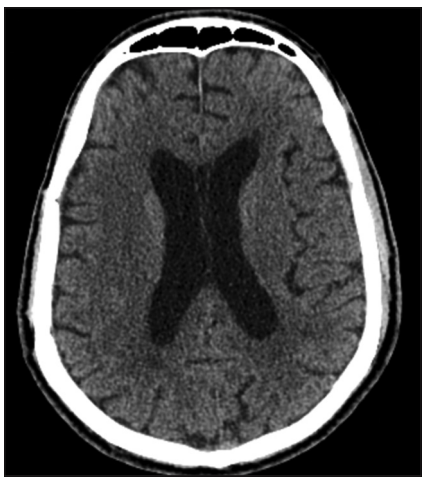

Figure 4: Follow up CT after 9 months

Due to CSF leakage in the spine, the brain sags, leading to the formation of SDH.[2] Younger patients with no history of trauma who present with SDH should raise suspicion for SIH.[2] In our case report, the patient was 53 years old with no traumatic history and a history of a thoracic epidural catheter. Known risk factors for SIH include hereditary connective tissue disorders such as Marfan syndrome, Ehlers-Danlos syndrome, and adult polycystic kidney diseases.[7]

Various diagnostic modalities, both invasive and non-invasive, are available. Non-invasive methods like MRI and CT are preferred, as invasive diagnostic modalities such as myelography can predispose patients to spinal CSF leaks.[2] Typical MRI findings in SIH include subdural fluid or blood collection, enhancement of pachymeninges, engorgement of venous structures, pituitary hyperemia, and brain sagging, collectively referred to as SEEPS.[2] While tonsillar herniation may occur due to low CSF and brain sagging, a study involving 56 SIH patients with CSF leaks showed that the average cerebellar tonsillar herniation was 5.5 mm above McRae's line, not below.[2] A nine-point brain MRI-based score has been proposed for SIH diagnosis, incorporating major and minor signs.[2] Major signs include pachymeningeal enhancement, venous sinus engorgement, and suprasellar cistern effacement, while minor signs include subdural collection, prepontine cistern effacement, and reduced mammilo-pontine distance.[2] A score exceeding 5 indicates a likelihood of CSF leak, with a specificity of 81.8% and a sensitivity of 88.9%.[2] Non-contrast whole-spine MRI is important for localizing the CSF leak in the spine and determining its anatomical pathology.[2]

Chronic SDH is a common condition in the elderly.[3] and a frequent sequel of SIH in younger patients. Studies have shown that many SIH patients with SDH and spinal CSF leaks can be treated with EBP. However, some patients may require multiple treatment modalities, including medical and surgical approaches. Surgical evacuation of the SDH is strongly indicated if the patient presents with neurological deficits or significant brain compression with midline shift. In cases without significant deficits and favorable imaging, conservative management, or EBP, is appropriate. Conservative management includes bed rest, oral hydration, caffeine, and steroids.[6] Prolonged bed rest is essential in the treatment regimen, but there is no established standard for the duration, as it varies among patients.[6] In our patient's case, bed rest for 48 hours followed by gradual head elevation was initially attempted, but an orthostatic headache recurred. Therefore, complete bed rest for 1 week was prescribed following the final targeted EBP.

EBP is an invasive technique in which 20–30 mL of venous blood is injected into the epidural space. There are primarily two types of EBP: targeted and blind. Targeted EBP, which identifies the source of the CSF leak, is more successful compared to blind EBP.[6] Targeted EBP typically involves identifying the source of the leak, which is usually in the thoracic region.[6] A study demonstrated a clinical improvement rate of 87.1% with targeted EBP versus 52% with blind EBP.[6]

Tranexamic acid has shown promising results in the treatment of chronic SDH.[3] It reduces plasmin activity by reversibly binding to lysine sites on plasminogen, thereby reducing fibrinolysis and inflammation.[3] Tranexamic acid can disrupt the cycle of neovascularization and neomembrane formation. It is generally safe and does not appear to increase the risk of venous thromboembolism, as indicated by the CRASH 2 and CRASH 3 trials for head injuries.[3] The ongoing TRACS and TORCH trials are investigating the use of tranexamic acid in chronic SDH and appear to be in favor of its use.[9]

The current mainstream surgical option for SIH is an EBP, and if it fails, open dural repair is considered.[7] Sobczyk *et al*. mention dural repair for SIH but emphasize that SIH treatment should be tailored to individual patients. Ahmad *et al*. concluded that SIH with chronic SDH requires a step-wise approach, as the majority of patients may improve with EBP and bed rest, although the duration of bed rest can be variable.[8]

CONCLUSION

- Clinical history and examination play a crucial role in the early diagnosis of SIH, especially the nature of headaches and associated symptoms.
- Prolonged bed rest with hydration is an essential measure for treating SIH, as many patients respond positively to these simple measures.
- EBP is the primary treatment modality for SIH, with targeted EBP being more effective than blind EBP. SIH with chronic SDH requires a step-wise approach, with consideration of burr hole evacuation for patients with significant neurological deficits and mass effects.
- Tranexamic acid has shown promising results in the treatment of chronic SDH, but long-term complications should be carefully evaluated.

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How to cite this article: Pattar RG, Karmarkar VS, Desai M. Spontaneous Intracranial Hypotension with Chronic Subdural Hematoma-A Case Report of Multimodality Management. Bombay Hosp J 2023;65(3):17-20.

Source of support: Nil, **Conflicts of interest:** None