Spontaneous intracranial hypotension secondary to anterior thoracic osteophyte: Resolution after primary dural repair via posterior approach

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ABSTRACT

INTRODUCTION: Spontaneous intracranial hypotension (SIH) is an uncommon syndrome widely attributed to CSF hypovolemia, typically secondary to spontaneous CSF leak. Although commonly associated with postural headache and variable neurological symptoms, one of the most severe consequences of SIH is bilateral subdural hematomas with resultant neurological deterioration.

PRESENTATION OF CASE: We present the case of a patient diagnosed with SIH secondary to an anteriorly positioned thoracic osteophyte with resultant dural disruption, who after multiple attempts at nonsurgical management developed bilateral subdural hematomas necessitating emergent surgical intervention. The patient underwent a unilateral posterior repair of his osteophyte with successful anterior decompression. At 36 months follow up, the patient reported completely resolved headaches with no focal neurological deficits.

DISCUSSION: We outline our posterior approach to repair of the dural defect and review the management algorithm for the treatment of patients with SIH. We also examine the current hypotheses as to the origin, pathophysiology, diagnosis and treatment of this syndrome.

CONCLUSION: A posterior approach was utilized to repair the dural defect caused by an anterior thoracic osteophyte in a patient with severe SIH complicated by bilateral subdural hematomas. This approach minimizes morbidity compared to an anterior approach and allowed for removal of the osteophyte and repair of the dural defect.

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1. Introduction

Spontaneous intracranial hypotension (SIH) is attributed to cerebrospinal fluid (CSF) volume depletion through spinal dural defects. Dural defects may be secondary to traumatic or degenerative spinal pathology.

Although SIH is commonly associated with postural headache and a variable constellation of neurological symptoms, the most severe consequence is subdural hematoma with resultant neurological devastation. Emergent neurosurgical evacuation is required, after which direct repair of dural defect or open placement of epidural blood patches (EBP) is warranted.

Five cases have been reported in which ventral vertebral osteophytes compromise dural integrity and lead to spontaneous CSF leakage and symptomatic SIH.1–5 In this article, we report a case of severe SIH complicated by bilateral subdural hematomas that was caused by an anterior thoracic osteophyte. The dural injury was repaired via a posterior surgical approach that we believe conferred lower morbidity.

2. Presentation of case

2.1. History and examination

The patient is a 36-year-old left-handed man who initially presented with severe positional headache, blurry vision, and tinnitus. He was discharged with a diagnosis of benign positional vertigo but subsequently presented to the emergency room with worsening symptoms. On MRI a non-enhancing, crescentic, extraxial mass was visualized in the anterior right temporal lobe (Fig. 1A). A lumbar puncture was unsuccessful under fluoroscopy. The patient improved on bed rest and hydration and was discharged with a diagnosis of SIH.

The patient returned four weeks later with persistent headache and bilateral tinnitus. His neurological exam was significant for left lateral gaze palsy and horizontal diplopia. Nonsurgical management with intravenous caffeine, as well as non-directed EBP at levels T7–8 and L1–2, were unsuccessful.

Further work-up with CT myelogram demonstrated a ventral epidural contrast collection (Fig. 2A and B). An isolated, sharp, and rostrally directed dorsal osteophyotic spur at the T1–2 level causing deformity of the ventral thecal sac was identified (Fig. 2A and B). An MRI with intrathecal gadolinium demonstrated extensive ventral epidural contrast collection and extravasation in adjacent soft tissue at T1–2, thereby confirming this as the site of CSF leak (Fig. 2C).
A directed EBP was applied at the T1–2 junction and the patient was discharged home after symptomatic improvement.

Eleven days following discharge, the patient presented again with altered mental status, severe 10/10 headache, and dysarthria. His condition deteriorated in the ED and neurological exam was significant for GCS 12, roving eye movements without tracking, lack of response to voice, and asymmetrical pupils. A head CT demonstrated large bilateral holohemispheric subdural collections with downward transtentorial herniation and effacement of the basilar cistern.

2.2. Operation and pathological findings

The patient was taken emergently for bilateral frontoparietal craniotomies to evacuate his subdural hematomas. CT head 24 h after surgical evacuation demonstrated recurrent, left greater than right holohemispheric subdural hematomas with adjacent mass effect and midline shift of 7 mm. Intrathecal saline infusion via lumbar drain was administered. Repeat CT on post-operative day seven demonstrated interval decrease in size of subdural collections.

On post-operative day eight, the patient underwent a unilateral posterior repair of his anterior thoracic osteophyte to prevent further complications of SIH. A hemilaminectomy at the T1/2 junction was performed, with resection of the T1 inferior facet and the T2 superior facet. A partial corpectomy allowed for adequate visualization of the anterior thoracic spine. This partial corpectomy and facetectomy allowed for a lateral approach to the nearly mid-line boney spur. Neuroradiological confirmed no changes in SSEPS or motor evoked potentials during the entirety of the procedure. A 5 mm ventral dural defect with associated arachnoid herniation was found at the level of the thoracic osteophyte. The osteophyte was readily palpated and resected using a combination of a Woodson elevator, downgoing curettes, and Penfield instruments. To address the dural defect, DuraGen in combination with morcellized Surgicel and fat graft was placed anteriorly. DuraGen was placed in a tack-up fashion and subsequently sealed with Tisseel. Posterior arthrodysis was then performed due to concern for progressive junctional kyphosis.

2.3. Postoperative course

Subsequent imaging confirmed removal of the osteophyte with successful anterior decompression and repair of the dural defect (Fig. 3). The patient was discharged home on post-operative day twelve after an uneventful ICU course. At 36 months follow up, the patient reported completely resolved headaches with no focal neurological deficits. He was able to return to work and resume his regular activities. The only complaint was occasional tinnitus and lightheadedness upon standing.

3. Discussion

3.1. Symptoms and presentation

SIH is an uncommon cause of headache and may present in association with a wide constellation of neurological symptoms. Postural headache is common but certainly not universal. Retrospective analyses have demonstrated diplopia to be the strongest positive predictor of CSF leaks in suspected SIH. Less commonly reported are presentations without any headache, as well as trigeminal neuralgia, syringomyelia, and acute cervical radiculopathies.

Unmanaged SIH may have unfavorable outcome including bilateral subdural hematomas and inferior displacement of posterior fossa elements. Thus, physicians treating suspected SIH must remain vigilant for development of potentially lethal complications, particularly in the elderly or those on anticoagulation who are more at risk for intracranial hemorrhage.
3.2. Diagnosis and evaluation

Diagnosis of SIH is made on the basis of symptoms, history, and MR findings. Although lumbar puncture is generally unnecessary, a CSF opening pressure of less than 60 cm of water is a classic finding. MR findings of pachymeningeal enhancement, venous engorgement, pseudo-subarachnoid hemorrhage, pituitary hyperemia, and/or cerebellar tonsil “sagging” (Fig. 1A) are helpful diagnostic features. Spinal imaging is generally reserved for patients with SIH who do not respond to non-directed EBP. Precise localization of the leak is best accomplished with CT myelography, though there is increasing interest in gadolinium myelography. In our case, high resolution CT myelography not only confirmed CSF extravasation but also identified the causative osteophyte for the dural defect.

3.3. Non-surgical therapy

Bed rest and hydration, intravenous caffeine and theophylline, as well as steroid therapy, have all provided temporary relief as part of conservative treatment regimens. EBP, which can provide symptomatic relief in as short as 2 h, often serve as first line therapy for CSF leaks of spontaneous or iatrogenic etiology. In the setting of failed patch attempt, reducing CSF pressures with acetazolamide and the use of reverse Trendelenburg position has been shown to enhance success.

In refractory leaks, efforts should be made towards specific localization of the leak followed by directed therapy, such as CT-guided percutaneous epidural injection of fibrin sealant, which is especially useful for excellent localization of large meningeal diverticula or tears.

3.4. Neurosurgical management

The surgical algorithm involves ligation or repair of overt diverticula and tears, or packing of the epidural space with a hemostatic agent when the source of the leak is not apparent. Dural injury by spinal osteophytes or disc herniation is rarely reported. Surgical treatment of CSF leaks caused by spinal osteophytes includes removal of the offending bone spur with surgical tamponade of the dural tear. Anterior approaches, as well as non-invasive EBP, have been reported. In our patient, a posterior approach was chosen to decrease morbidity.

In general, the posterior approach is indicated in patients with acute comorbid pathologies such as widespread malignancy, spinal cord compression, or poor tolerance to the anterior approach. However, in patients with pathology confined to anterior elements and who do not require posterior instrumentation, such as ours, there is no clear contraindication to anterior access. The benefits of anterior exposure, namely excellent visualization, have to be balanced against the risk of greater soft tissue damage and extended length of the operation. In our case, given the patient’s low-normal BMI, youthful boney elements, and localized target, we were confident adequate exposure could be obtained via the posterior approach. In addition, the posterior angle allowed for the tacking of the dural repair to tamponade the source of the CSF leak since primary closure was not possible.

Furthermore, acute neurological decompensation secondary to expanding subdural hematoma requires immediate surgical intervention. In our patient, although evacuation was initially successful, the hematomas recurred on a smaller scale. Continuous intrathecal saline infusion via lumbar drain has been used to directly augment intracranial pressure by replacing lost fluid, and has been reported in the setting of severe SIH to aid in restoring physiology CSF volume and flow.

4. Conclusion

We present a posterior approach for repair of a dural defect caused by an anterior thoracic osteophyte in a patient with severe SIH complicated by bilateral subdural hematomas. This approach minimizes morbidity compared to an anterior approach and allowed for removal of the osteophyte and repair of the dural defect.

Conflict of interest

None.

Funding

None.

Ethical approval

Written consent has been obtained and approved from IRB and the patient.

Author contributions

AV, GG, and BJ contributed to the drafting of the manuscript. SCB provided critical revision. SDC and SAM conceived of the project and performed the surgical intervention and consented the patient.
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