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Spontaneous Intracranial Hypotension Secondary to Spondylotic Spine Osteophyte

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ABSTRACT

Background and Objective: *Orthostatic headache is the key symptom of intracranial hypotension secondary to cerebral spinal fluid (CSF) leak. Spontaneous intracranial hypotension (SIH), i.e. intracranial hypotension in the absence of a known iatrogenic dural puncture, is an under recognized cause of orthostatic headache caused by intracranial hypotension. The most common cause of SIH is thought to be leakage of CSF from ruptured perineural cysts (i.e. Tarlov cysts). Conservative treatment, epidural blood patch (EBP) procedure and open surgical repair have been used to treat SIH.*

Case Report: *Here we report a case of patient with SIH who was initially treated with epidural blood patches with short term symptomatic relief when the CSF leakage site was not identified, but later developed recurrent headache with rapidly declining mental status and severe neurological symptoms. A dural tear related to a spondylotic osteophyte and life threatening brain herniation was identified with additional imaging studies. The patient eventually required urgent surgical repair with favorable clinical outcome.*

Conclusion: *This and other cases of SIH caused by spondylotic osteophyte-related durotomy suggest that SIH caused by spondylotic osteophyte has a different clinical course than other SIH cases. Such patients may only have transient response to epidural blood patch; their symptoms may acutely worsen due to larger durotomy by spondylotic osteophytes. Clinicians should be vigilant in monitoring neurological symptoms and consider spondylotic osteophyte as a potential etiology of spontaneous dural puncture. Once such etiology is identified, early surgical repair is indicated.*

Keywords

Spontaneous intracranial hypotension, Epidural blood patch, Spinal osteophytes.

Introduction

Positional headache is a hallmark of post dural puncture headache (PDPH) which is a relatively common iatrogenic condition resulting from intentional or unintentional puncture of dura during procedures involving intrathecal or epidural space access such as lumbar puncture, spinal anesthesia for the former and epidural catheter placement or epidural steroid injections for the latter [1]. PDPH occurs when a slow leak of cerebrospinal fluid leads to contraction of the subarachnoid space and compensatory expansion of the pain-sensitive intracerebral veins [2]. The same positional headache and associated nausea, vomiting, diplopia dizziness and tinnitus may also present in the absence of a known dural puncture event – a clinical entity that has been termed spontaneous intracranial hypotension (SIH) [3,4]. The most common cause of SIH is CSF leakage from a spinal meningeal diverticulum, perineural cyst (Tarlov cysts) or simple dural tear [5]. The clinical outcome of SIH has been thought to be largely favorable, with most patients either have spontaneous recovery [6] or resolution after epidural blood patch (EBP) procedures [3]. Here we present a case of SIH secondary to durotomy caused by a spondylotic osteophyte who was treated initially with EBP but developed life threatening deterioration necessitating urgent surgical intervention, suggesting

that SIH associate with osteophyte related dual tear may need close monitoring and prompt intervention.

Clinical Case

The patient was a 58 y/o otherwise healthy male who presented to emergency department with 2 weeks of bi-frontal headache and fatigue, and 1 week of progressive horizontal diplopia. Symptoms first started after intercourse that was described "very active". The headache was positional in nature, improved while lying down and became worse with sitting up. He has had no recent spine procedures. His past medical history and surgical history is significant for lumbar discectomy for lumbar disk herniation 15 years ago.

At initial presentation, he had no associated symptoms of nausea or vomiting, no fever or chills. Physical exam was unremarkable. A lumbar puncture was performed for work up of potential infectious etiology. During the lumbar puncture a low opening pressure of 6 cm H2O was noted. Cerebrospinal fluid cell count and chemistry were within normal limits. His positional headache was exacerbated by the lumbar puncture. Brain magnetic resonance imaging (MRI) showed diffuse, smooth pachymeningeal enhancement consistent with intracranial hypotension (Figure 1). MRI of cervical, thoracic and lumbar spine with FIESTA (Fast Imaging Employing Steady-state Acquisition) protocol showed multilevel degenerative changes throughout the spine without identification of CSF leak (Figure 2). A diagnosis of SIH was established.

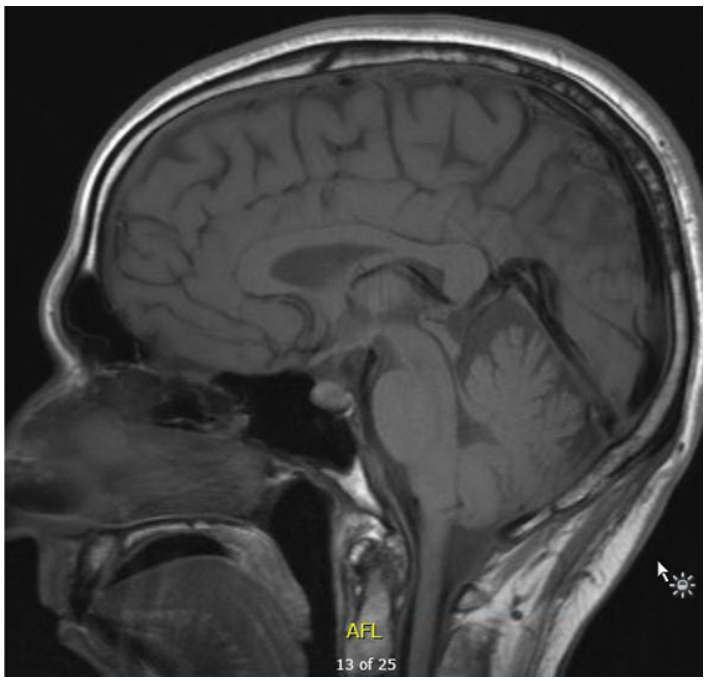

Figure 1: Brain MRI on Initial presentation: Diffuse, smooth pachymeningeal enhancement consistent with intracranial hypotension on T2 weighted image.

Subsequently, a fluoroscopy-guided EBP procedure was performed at L3/4 interlaminar space with 20cc of autologous blood. His headache improved after the procedure but recurred after one week.

A second fluoroscopy-guided EBP was performed approximately 3 weeks after the initial EBP. Because of the transient benefit from initial lumbar epidural blood patch, decision was made to perform the second EBP at T12/L1 level with 20 cc of autologous blood under the hypothesis that a blood patch procedure at a higher site in the spine may be able to cover an unidentified small CSF leak at the thoracic or thoracolumbar junction level. The patient again experienced transient improvement of his positional headache but not the diplopia for approximately one week.

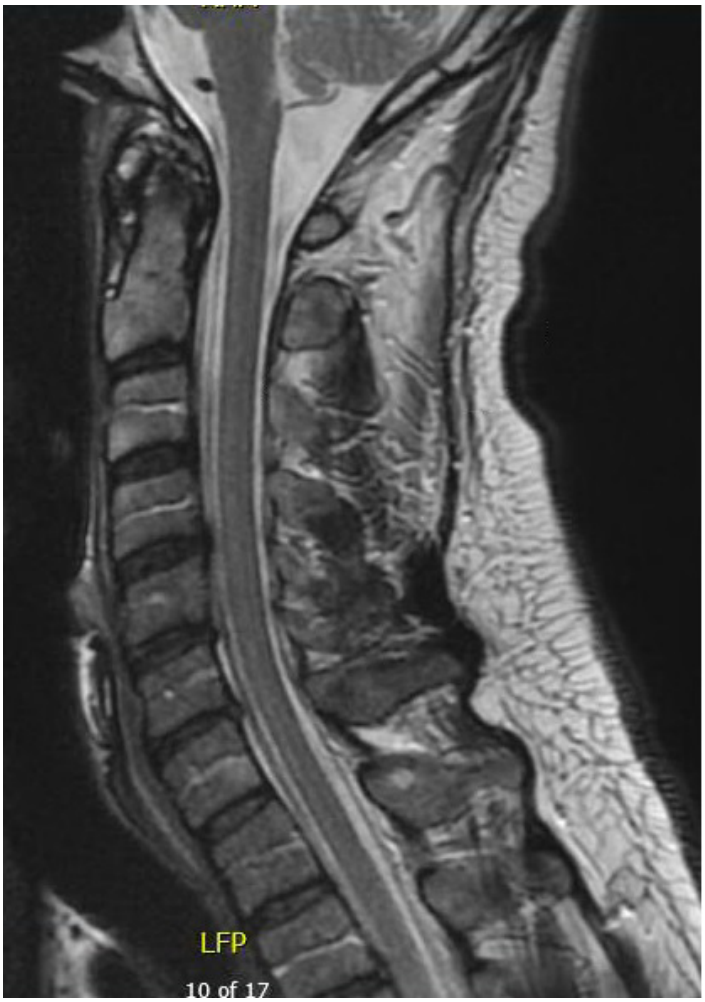

Figure 2: Cervical spine MRI on initial presentation. On T2 weighted image, a posterior disc-osteophyte complex was seen at C6-7 level. No CSF collection in ventral or dorsal epidural space was detected.

A CT myelogram was then planned to further identify leakage site for more targeted intervention. While awaiting CT myelogram the patient had steady decline with worsening headache and diplopia and was admitted in Emergency Department for observation. He developed intermittent altered mental status in the next 24 hours. Repeat brain MRI showed new bilateral convexity subdural fluid collections. Spine MRI showed thin dorsal extradural collection extending from the level C3-C4 through T5-T6 (Figure 3), suggesting a potential CSF leakage site between C3 and T6. A third fluoroscopic guided epidural blood patches procedure at T2-3 interlaminar space level was performed with 20 cc of autologous

blood. Patient tolerated the procedure well; there were no new neurological symptoms after the EBP procedure.

His symptoms again improved for the next 36 hours, however, he developed acutely declining mental status and upgaze palsy, and he was noted to have increasing confusion and developed a stuporous-like condition and was admitted to ICU for decreased arousal. Urgent brain CT showed progressive enlargement of bilateral subdural hematomas and bilateral brain uncal herniation (Figure 4). He was taken to the OR for urgent bilateral burr hole subdural drain placement. Subsequently, a CT myelogram was performed which revealed epidural contrast collection from C3 to T5, consistent with CSF leak. There was ventral epidural collection at C7 as a suspicious source of the CSF leak, most likely related to prominent small right paracentral spiculated calcified disc/osteophyte at the C6-7 level (Figure 5). He underwent C6-C7 discectomy, removal of osteophyte and instrumented fusion. Intra-operatively, a dural opening just to the right of the midline along the C6-C7 disc space was noted. Active CSF leakage from ventral dural sac was observed. This dural opening was repaired with fibrin glue and a piece of harvested autologous platysma muscle. Post operatively, his mental status improved quickly. Headache resolved. He was discharged to acute inpatient rehabilitation and then home after a 2-week rehabilitation course. His diplopia resolved in the next month. At 2 month and 6 month follow up, he remained symptom free and returned to baseline function.

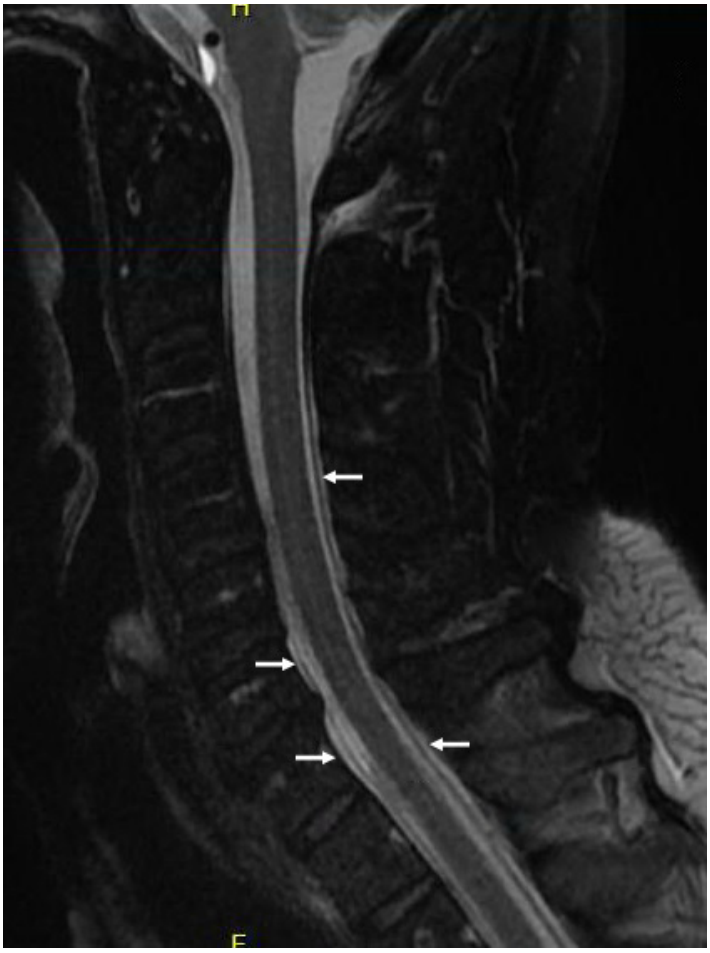

Figure 3: Repeat cervical Spine MRI after patient presented with recurring headache and worsening clinical symptoms. Long segment, thin dorsal extradural collection extending from the level C3-C4 through T5-T6 was seen on T2 weighted images (arrows), consistent with a CSF leak.

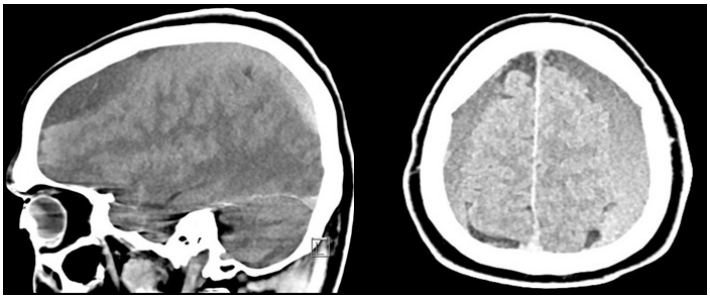

Figure 4: Brain CT after the patient had acute worsening mental status change showing bilateral subdural hematomas with mass effect, uncal herniation.

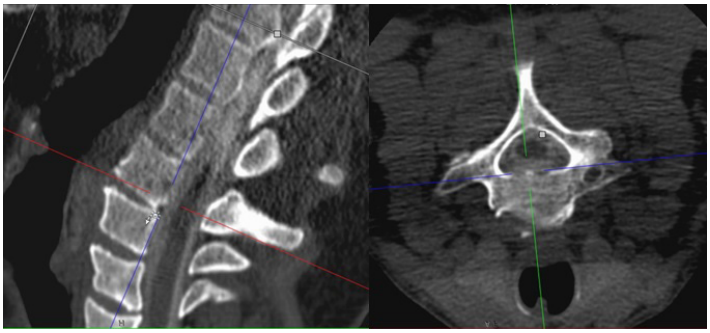

Figure 5: CT myelogram shows ventral collection of contrast at C7 in a configuration most consistent with extravasation into the epidural space. This is adjacent to a small right paracentral slightly spiculated calcified disc or osteophyte at the C6-7 level.

Discussion

SIH is a relatively rare and under-recognized condition with an estimated prevalence and incidence of are 1:50,000 and 5:100,000, respectively [4]. Spontaneous leakage of CSF is believed to be the cause of intracranial hypotension. Etiology and sources of spontaneous leakage of CSF in this condition is not completely understood. The most common cause of SIH is CSF leakage from a spinal meningeal diverticulum, perineural cyst (Tarlov cysts) or simple dural tear [5]. Connective tissue abnormality such as Marfan's syndrome has been shown to be associated with IIH [7,8]. A few cases of spinal osteophytes causing spontaneous dura puncture and intracranial hypotension have also been reported [9-15]. MRI and CT myelogram are the two most commonly employed imaging tests to identify site of CSF leakages [16]. Radionucleotide cisternography can also be used but has a high false negative rate (30%) [4]. MR myelography may offer higher sensitivity than CT myelography [16]. Most spinal CSF leaks are located at the cervicothoracic junction or in the thoracic spine, and they may be associated with meningeal diverticula [4,16].

The clinical course of SIH is in general benign and has a favorable response to EBP. In a retrospective analysis of 74 patients who underwent EBP procedures, Angelo et all reported that 87% of patients returned to normal life in a few weeks with resolution of

symptoms; none of their patients needed open surgical repair of the CSF leak [3]. There are also case reports that symptoms from SIH can resolve with no intervention [17,18]. However, this may not be the case for SIH that is caused by spinal osteophytes. A review of all reported cases of SIH secondary to spinal osteophytes [9-15] showed that all but one [10] patient had inadequate responses to EBP procedures and required surgical removal of osteophytes and dural repair. Outcome after surgical intervention was favorable in all cases [9,11-15].

Our case is notably different from previously reported cases with regards to the difficulty in identifying a leakage site initially and the rapid decline of the patient's clinical condition. In this case, a definite site of CSF leakage and CSF collection in the epidural space could not be identified in the initial presentation. Empirical EBP at lumbar and thoracolumbar junction levels could only provide short term benefit. CSF collection and leakage site was only identified when patient presented with acute worsening symptoms, at which point a fluoroscopic guided EBP was performed close to the leakage site but only with only transient benefit. The patient's symptoms acutely declined with life threatening brain herniation, which required urgent surgical intervention.

It is likely that initially the dural tear was small and the loss of CSF was slow. Even though the patient was symptomatic, the slow CSF was not detectable with MRI. Patient was treated empirically with EBP procedures with a total volume of 60 cc spanning 6 weeks, all of which provided only transient symptomatic improvement. It is unclear to us whether the EBP procedures might have contributed to the rapid deterioration of the patient's clinical course. Conceptually, a large volume of blood administered into the dorsal epidural space, particularly close to the dural tear location, may displace the spinal sac anteriorly against the osteophyte located ventral/anterior to the dural sac and has the potential to exacerbate the dural tear. While a causal relationship cannot be established with a single case report, clinicians should consider this possibility and carefully weigh the benefit and risk of EBP when considering treatment of osteophyte related SIH. Further studies of similar cases are needed to delineate the optimal management in this condition.

This case and other reported cases highlight spondylotic osteophyte as a cause of spontaneous dural puncture, which may be more common than previously thought. This subtype of SIH has a different clinical course in that it is less likely to respond to conservative therapy and EBP procedure. Its clinical course can fluctuate and acutely deteriorate. Surgical repair is indicated once osteophyte causing dural tear is identified to avoid potential worsening of symptoms and life threatening secondary brain herniation. In patients with SIH for which a site of CSF leakage cannot be identified, clinician should be vigilant of potential sudden worsening of symptoms and development of life threatening brain herniation due to precipitately decreasing intracranial pressure. The risk of repeat epidural blood patch procedure should be carefully considered. In such scenario, emergent MRI and CT myelogram are indicated to identify large dural leak and surgical repair is warranted.

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