

Intracranial hypotension due to ventral thoracic dural tear secondary to osteophyte complex: resolution after transdural thoracic microdiscectomy with dural repair. Illustrative case

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BACKGROUND Intracranial hypotension (IH) manifests with orthostatic headaches secondary to cerebrospinal fluid (CSF) hypovolemia. Common iatrogenic etiologies include lumbar punctures and spinal surgery. Although much rarer, structural defects such as osteophytes and herniated calcified discs can violate dural integrity, resulting in CSF leak.

OBSERVATIONS The authors reported the case of a 32-year-old woman who presented with progressively worsening orthostatic headaches. During an extensive examination, magnetic resonance imaging of her thoracic spine revealed a cervicothoracic ventral epidural collection of CSF, prompting a dynamic computed tomography myelogram, which not only helped to confirm severe cerebral hypotension but also suggested underlying pathology of a dorsally projecting disc osteophyte complex at T2–3. Conservative and medical management failed to alleviate symptoms, and a permanent surgical cure was eventually sought. The patient underwent a transdural thoracic discectomy with dural repair, which resulted in resolution of her symptoms.

LESSONS Clear guidelines regarding the management strategy of IH secondary to disc osteophyte complexes are yet to be established. A thorough literature review noted only 24 reported cases between 1998 and 2019, in which 13 patients received surgery. There is a 46% symptom resolution rate with conservative management, lower than that for iatrogenic etiologies. For patients in whom conservative management failed, surgical intervention proved effective in resolving symptoms, with a success rate of 92.3%.

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KEYWORDS intracranial hypotension; orthostatic headache; osteophyte; cerebrospinal fluid leak; blood patch; thoracic discectomy

Intracranial hypotension (IH) manifests as positional headaches due to cerebrospinal fluid (CSF) hypovolemia, with other symptoms including nausea, diplopia, tinnitus, neck stiffness, and photophobia.^{1,2} Radiographic signs include pachymeningeal enhancement, subdural effusions or chronic hemorrhages, pituitary engorgement, and venous distention. Acquired tonsillar ectopia can lead IH to be confused for type 1 Chiari malformation, although there are multiple measures of midbrain sag to facilitate discrimination between the two.³ CSF leaks can be idiopathic or may occur iatrogenically after a lumbar puncture or spinal procedure. Although rarer, structural

defects in the spinal column, such as severe degenerative bony pathology (e.g., calcified disc osteophyte complexes) can tear the dura and precipitate a CSF leak.

The incidence of IH is estimated at 5 per 100,000 patients, most common in thin women who suffer from a connective tissue disorder (CTD) such as Marfan syndrome, Ehlers-Danlos syndrome (EDS) type 2, or autosomal dominant polycystic kidney disease.^{2,4} The initial approach is often conservative; bed rest, saline infusions, and empirical autologous blood patches may provide rapid relief in up to 90% of patients.^{4,5} If these approaches fail, targeted, interventional

ABBREVIATIONS CSF = cerebrospinal fluid; CT = computed tomography; CTD = connective tissue disorder; EBP = epidural blood patch; EDS = Ehlers-Danlos syndrome; ICHD = International Classification of Headache Disorders; IH = intracranial hypotension; MRI = magnetic resonance imaging.

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radiology-guided blood patches and surgical management options are subsequently considered.

Illustrative Case

A 32-year-old right-handed woman (body mass index 17.8) with no significant medical, surgical, or trauma history presented with a 1-month history of progressively worsening bifrontal and occipital headaches. These headaches were exacerbated with activity, coughing, sneezing, standing, and Valsalva maneuvers. The patient had accompanying constant dull and occasionally shooting neck and back pain, which also worsened with activity and was relieved with rest. A trial of conservative management with Fioricet (acetaminophen/butalbital/caffeine) and Sumatriptan failed. The pain interfered with activities of daily living, including the inability to tolerate sitting for >30 minutes, standing for >10 minutes, or walking >1 block. Underlying CTDs such as EDS were ruled out. Family history was noncontributory. Review of symptoms revealed muffled hearing, right-sided tinnitus, and otalgia, however, visual symptoms were absent. Of note, the musculoskeletal, spinal, and neurological examination results were within normal limits. The initial differential diagnosis included migraine, Chiari malformation and space-occupying lesions, subarachnoid hemorrhage, cerebral venous sinus thrombosis, CSF fistula, and occult CSF leak.

Investigations and Conservative Management

Preliminary imaging (magnetic resonance imaging [MRI], MR angiography, and MR venography) results were negative for cerebellar sag, subdural collections, venous distention, and pachymeningeal enhancement on initial review. The patient's neck pain was investigated with an MRI, which showed a ventral epidural collection that extended past the cervicothoracic junction. A consequent MRI of the thoracic and lumbar spine demonstrated a large circumferential epidural fluid collection matching CSF signal intensity throughout the cervical, thoracic, and upper lumbar spine. Moreover, moderate thecal sac narrowing from T2 to T5 due to the epidural collection was noted (Fig. 1). A nonfocal lumbar epidural blood patch (EBP) (~7 mL) was performed with no improvement in symptoms. Our patient then received a second nonfocal EBP (12 mL) on the same day that resulted in mild improvement of her symptoms. After discharge, Fioricet, taken as needed, and nonsteroidal antiinflammatory drugs were continued with minimal relief. Further outpatient examination with a dynamic CT myelogram of the spine revealed an opening pressure of 14 mm Hg and a robust high-flow CSF leak into the ventral epidural space extending cranial, likely secondary to a dorsally projecting complex of small osteophytes at levels T2–3 eroding through the dura (Fig. 2). Delayed images demonstrated progressive accumulation of epidural contrast consistent with CSF leak. She received CT-guided T2–3 EBP. Once again, an overall mild improvement in symptom severity was noted before the return of symptoms to baseline. At that point, a permanent surgical solution was considered.

Surgery and Postoperative Course

The patient received laminectomies at T2–3 to allow for a unilateral transection of the dentate ligament to mobilize the cord, thus allowing for a ventral dural exploration. Upon mobilization of the cord, a clear 1.5-cm vertical erosion in the ventral dura running rostral-caudal directly in the midline, exposing the ventral disc osteophyte complex responsible for this tear (Fig. 3A). A transdural discectomy was performed (Fig. 3B). The disc osteophyte complex specimen measuring



FIG. 1. Preoperative T2 sagittal MRI spine demonstrating ventral and dorsal epidural CSF collection extending throughout the cervical, thoracic, and upper lumbar spine (arrows).

$0.3 \times 0.2 \times 0.2$ cm was sent to pathology for analysis and returned as paucicellular tissue and calcific debris. After a primary repair of the dural defect with sutures (Fig. 3C), a ventral sling of bovine pericardium was used as a supplemental barrier between the spinal cord and the repaired surface of the dura (Fig. 3D) (Video 1). Postoperative lumbar drainage was continued for 3 subsequent days, and the patient was ultimately discharged home on postoperative day 5 with complete resolution of her symptoms and no complications. She remains headache free past her 1-year follow-up.

VIDEO 1. Clip showing intraoperative view of transdural thoracic discectomy followed by primary and secondary dural repair to achieve a watertight dural closure. [Click here to view.](#)

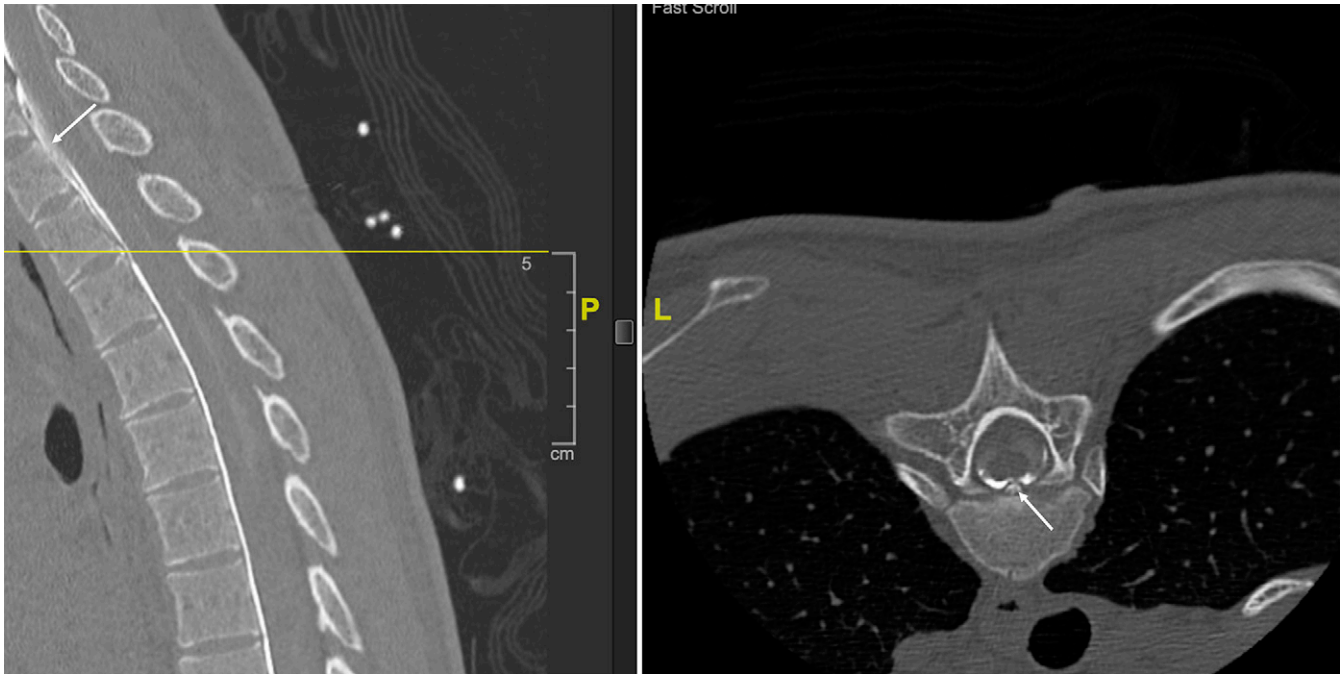


FIG. 2. Preoperative CT myelogram sagittal section (**left**). Arrow demonstrates ventral CSF epidural collection. Arrow on the axial section (**right**) shows the calcified disc osteophyte complex resulting in ventral dural invasion.

Literature Review

We searched the PubMed (Medline) and Embase databases in English language using a publication year ranging from 1970 to 2020 for “CSF leak OR Intracranial hypotension” AND “disc OR osteophyte OR disc-osteophyte complex.” Search results captured a total of 203 papers (n = 112 for PubMed and n = 91 for Embase). After excluding 50 duplicates, we were left with 153 papers. Of these, we selected the case reports and series that pertained to CSF leak secondary to structural defects and in line with our interest for collating a

comprehensive summary of all reported patient cases to date. This included 16 case reports (including one neuroimage) and 3 case series yielding a total of 24 reported cases (Fig. 4).

Discussion

The International Classification of Headache Disorders (ICHD-3) diagnostic criteria for headaches secondary to low CSF pressure include headaches with low opening pressures on diagnostic studies and/or radiographic evidence of a CSF leak, absence of

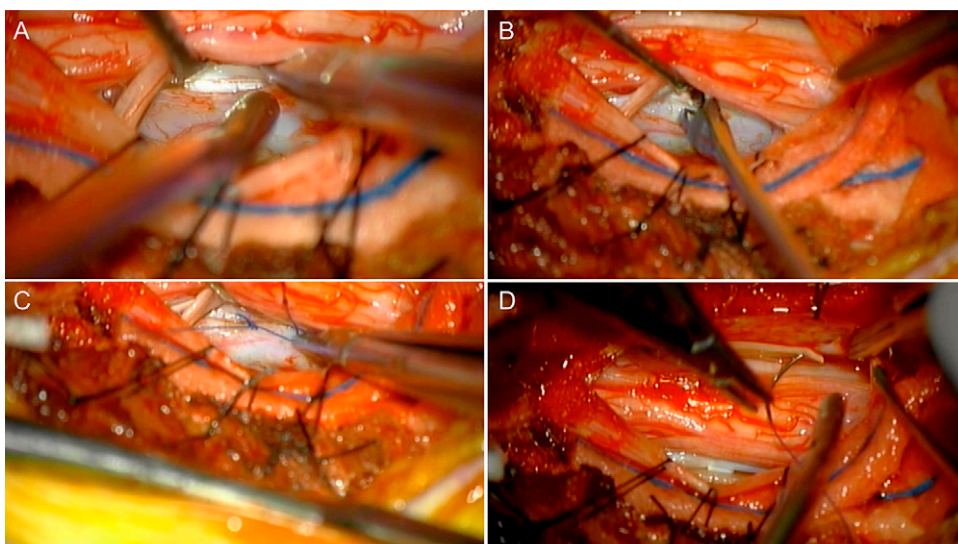


FIG. 3. Intraoperative images. **A:** Ventral tear in the dura prior to repair. **B:** Performing the transdural discectomy. **C:** Primary repair of the dura with 7-0 Prolene sutures. **D:** Secondary repair with a ventral sling of the bovine pericardium that was tacked to the inside of the dura at the four corners.

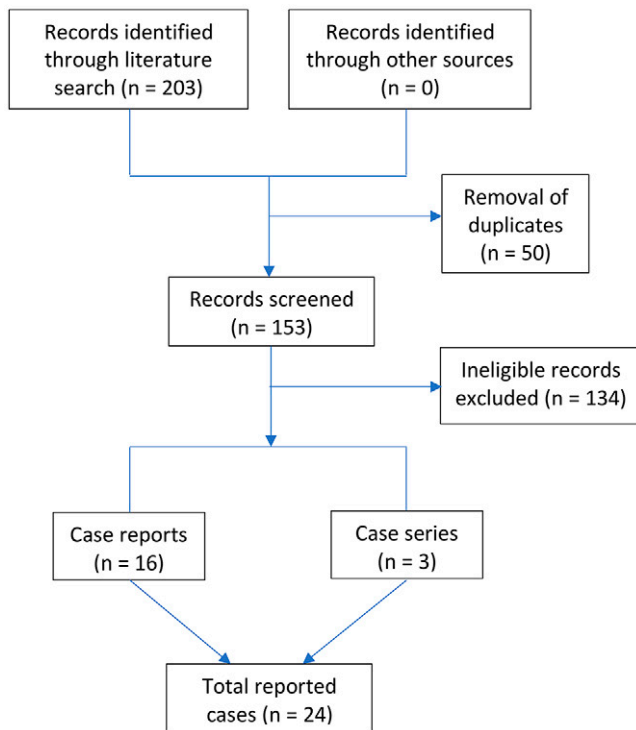


FIG. 4. Flow diagram showing literature database search and patient selection methodology.

procedure(s) or trauma that can cause a CSF leak, headaches that have occurred in the setting of low CSF pressure or CSF leakage that has led to its discovery, and headaches that cannot be accounted for by any other ICHD-3 diagnosis.⁶ IH manifests as postural headaches that worsen with activity and improve with lying supine, and they are often sudden in onset. Subarachnoid hemorrhage needs to be ruled out if history so suggests. Other commonly associated symptoms include nausea, vomiting, tinnitus, vertigo, and neck stiffness. Moreover, cranial nerve palsies secondary to sagging of the brain due to hypovolemia rarely occur, although they were not noted in our patient.^{7,8} In practice, orthostatic headaches along with either radiological evidence of CSF hypotension/CSF leak or opening lumbar pressure of <60 mm H₂O is sufficient for a diagnosis of IH.⁷ MRI of the brain is the primary diagnostic test for suspected intracranial hypotension. Typical cranial imaging features of CSF hypovolemia include subdural fluid collection and presence of extrathecal CSF, enhancement of the pachymeninges, engorgement of the venous structures, pituitary hyperemia, and downward displacement of the cerebellar tonsils.⁵ Rarely, MRI results are unrevealing as in the case of our patient, whose scan did not demonstrate any findings despite her opening pressure of 14 mm Hg. Most CSF leaks in the reported literature have occurred at the cervicothoracic junction or the thoracic spine.⁴ An MRI of the spine can help localize dural defects in some cases; however, in our case, the osteophyte complex at T2–3, which was suspected as the cause of dural erosion, was better evaluated on dynamic CT myelography, the gold standard for localizing leaks and guiding targeted EBPs or surgical planning.⁹

Observations

We provide an updated summary of all reported cases of dural tears secondary to disc osteophytes leading to IH. A thorough

review of the existing English literature between 1970 and 2020 revealed only 24 reported cases (Table 1).^{10–28} Osteophyte involvement was seen in 13 of the 24 cases. The diagnosis was more common in women (ratio 1.67:1), and the mean age at diagnosis was 41.8 years (range 25 to 57); 79.2% patients (n = 19) had pathology in the thoracic spine versus 20.8% patients (n = 5) with cervical spine pathology. Eleven patients (46%) received surgery after a failed trial of EBPs, and 2 patients (8%) received surgery before trial of EBPs. For the remaining 46% of patients (n = 11), the headache symptoms improved or resolved with conservative measures alone (including EBPs). Only 1 patient had persistent symptoms at follow-up (32 months) despite receiving surgery.¹¹ The remaining 12 surgical patients demonstrated improvement or resolution of their symptoms with an average of 14 months of follow-up. In our patient, symptom relief was not achieved despite medication management, two nonfocal EBPs, and one targeted EBP; thus, surgical management was considered as a last, curative resort. Surgery involves removal of the offending bony lesion osteophyte complex to avoid future recurrence of symptoms and dural reconstruction, which can be challenging given the ventral location and working around the spinal cord. Placement of a lumbar drain allows maintenance of low CSF pressures to allow time for the dura to reseal. Placement of a lumbar drain or a lumboperitoneal shunt was mentioned in only 3 of the 13 surgically managed patients according to our literature review.

Lessons

Conservative management of postprocedural CSF is successful in most cases. There is a success rate of 92.3% in patients who receive surgery, although surgical intervention involves a complex repair of the dural defect while working around the spinal cord in a limited area. Our literature review noted that some patients with CSF leaks secondary to a structural pathology improved with EBPs alone, although notably less than the improvement rate when a leak occurred iatrogenically, whereas others required surgical repair before symptom improvement/resolution was noted. (The reasons and factors for the same are unclear, and the literature does not report long-term follow-up data with regards to CSF leak recurrence in patients who experienced improvement or symptom resolution with conservative/medical management.) The number of EBPs may vary depending on the severity of symptoms, degree of impact on the quality of life, and patient and consultant preference. Clear guidelines regarding the management strategy of IH secondary to bony pathology are yet to be established.

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TABLE 1. Symptoms, management, and outcomes of patients with spontaneous intracranial hypotension caused by spinal osteophytes and disc herniation

Authors & Year	Age (yrs), Sex	Symptoms at Presentation	Spinal Disorder	EBP (B, T)	Surgery (Y/N)	Surgical Details	HA Outcome, FU
Vishteh et al., 1998 ¹⁰	32, M	HA	C5-6 osteophyte	1, 0	Y	Anterior discectomy (C5-6), Osteophylectomy, 1° dural repair (2× interrupted Prolene sutures)	Resolved, ND
Eross et al., 2002 ¹¹	44, F	HA, ear fullness, nasal tightness, paraesthesia, retro-orbital pressure	C5-6 osteophyte + disc	1, 2	Y	C5 discectomy, Partial C5-6 & C6-7 corpectomies, 1° dural repair, lumboperitoneal shunt placement	Remained, 32 mos
Eross et al., 2002 ¹¹	39, F	HA, neck stiffness, nausea, vomiting, sensitivity to changes in position	C7-T1 osteophyte + disc	3, 0	Y	Anterior cervical discectomy/osteophylectomy, fusion at C7-T1 with an allograft & metal plate, 1° dural repair, lumbar drain placement	Improved, 1 mo
Eross et al., 2002 ¹¹	46, F	HA, posterior neck pain, tinnitus, epistaxis	C4-5 osteophyte + disc	1, 0	N		Improved, 2 mos
Winter et al., 2002 ¹²	42, F	HA, nausea, dizziness, tinnitus	T7-8 calcified disc	0, 1	Y	Burr hole drainage of the subdural CSF collection ×2	Resolved, 18 mos
Rappoport et al., 2003 ¹³	37, F	HA, interscapular pain, nausea, photophobia	T7-8 disc	0, 2	N		Improved, ND
Binder et al., 2005 ¹⁴	55, F	HA, aural fullness, memory difficulties, disorientation	T2-3 osteophyte	1, 0	Y	Anterior discectomy (T2-3), osteophylectomy, 1° dural repair (2× nonabsorbable vertical mattress sutures & a small piece of interposed muscle → fibrin glue)	Resolved, 12 mos
Yokota et al., 2008 ¹⁵	25, F	HA, tinnitus	T8-9 osteophyte	1, 1	N		Resolved, 24 mos
Witiw et al., 2012 ¹⁶	46, F	HA, nausea, vomiting	C4-5 calcified disc	3, 0	Y	Anterior discectomy (C4-5), osteophylectomy, posterior longitudinal ligament resection, no primary closure (corpectomy deemed too invasive), DuraMatrix collagen dura substituted membrane inserted followed by Tisseel, Solis cervical cage packed w/ autologous bone placed in the intervertebral space	Resolved, 2 mos
Hasiloglu et al., 2012 ¹⁷	32, F	HA, neck pain & stiffness, nausea	T2-3 osteophyte + disc	1, 0	N		Resolved, 4 mos
Hasiloglu et al., 2012 ¹⁷	42, F	HA, dizziness, vertigo, nausea, vomiting	T5-6, T11-12 osteophytes + disc	0, 1	N		Resolved, 6 mos
Allmendinger & Lee, 2013 ¹⁸	56, F	HA	T6-7 herniated calcified disc	1, 1	N		Resolved, ND
Allmendinger & Lee, 2013 ¹⁸	47, M	HA, nausea	T6-7 calcified disc	0, 2	N		Resolved, ND

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TABLE 1. Symptoms, management, and outcomes of patients with spontaneous intracranial hypotension caused by spinal osteophytes and disc herniation

Authors & Year	Age (yrs), Sex	Symptoms at Presentation	Spinal Disorder	EBP (B, T)	Surgery (Y/N)	Surgical Details	HA Outcome, FU
Allmendinger & Lee, 2013 ¹⁸	49, M	HA	T5–6 osteophyte + disc	0, 2	N		Resolved, ND
Veeravagu et al., 2013 ¹⁹	36, M	HA, blurry vision, tinnitus	T1–2 osteophyte	1, 2	Y	Posterior hemilaminectomy (T1–2), partial corpectomy & facetectomy, osteophyctomy, DuraGen + morcellized Surgical & fat graft (placed anteriorly), DuraGen sealed w/ Tisseel	Resolved, 36 mos
Wilson et al., 2013 ²⁰	47, M	HA, reduced level of consciousness	T6–7 calcified disc	3, 0	Y	Hemilaminectomy & costotransversectomy (T6–7), CSF leak repaired w/ a muscle, fascia, & fat graft, secured w/ fibrin glue	Resolved, 6 mos
Agarwal et al., 2013 ²¹	57, M	HA	T6–7 calcified disc	0, 1	N		Resolved, 5 wks
Rapoport et al., 2014 ²²	45, M	HA, blurry vision	T12–L1 disc	0, 0	Y	Laminectomy (T12–L1), dura incised dorsally → removal of intradural disc fragment, 1° repair of the anterior dural defect	Resolved, 3 wks
Hung & Hsu, 2015 ²³	34, F	Postural HAs, neck tightness	T2–3 osteophyte	2, 1	N		Resolved, 20 mos
Pricola Fehnel & Borges, 2015 ²⁴	34, M	Postural HAs, bilateral shoulder discomfort, nausea, vomiting	T9–10 calcified disc	2, 0	Y	Posterior laminoplasty (T9–10), dorsal durotomy, transdural discectomy, closure of the ventral dural defect achieved by running 5-0 silk sutures, autologous fat graft (placed ventral to the durotomy & posterior to the disc space), Closure of the dorsal durotomy achieved w/ running 5-0 Prolene sutures followed by covering w/ a small autologous fat graft, T9–10 laminae replaced w/ a 2.0 titanium miniplate	Resolved, 1 mo
Dash et al., 2016 ²⁵	38, F	HA, nausea, vomiting, muffling noise in the ears, worsening gait	T12–L1 osteophyte	Multiple, not specified	Y	Hemilaminectomy (T12–L1), osteophyctomy, 1°, deemed not possible due to location of durotomy, DuraGen placed w/ addition of Tisseel, lumbar drain placement	Resolved, 4 yrs
Sattip et al., 2016 ²⁶	41, F	Positional HAs, photophobia, nausea, vomiting	T5–6 calcified disc	0, 2	N		Resolved, 9 yrs
Urbach et al., 2019 ²⁷	34, M	HA	T2–3 osteophyte	0, 0	Y	ND	Resolved, ND
Fiechter et al., 2019 ²⁸	46, F	HA	T12–L1 disc	3, 0	Y	Laminectomy (T12), intradural disc sequestration, watertight ventral & dorsal dural closure, dorsal dural closure & T12 laminoplasty	Resolved, 3 mos

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TABLE 1. Symptoms, management, and outcomes of patients with spontaneous intracranial hypotension caused by spinal osteophytes and disc herniation

Authors & Year	Age (yrs), Sex	Symptoms at Presentation	Spinal Disorder	EBP (B, T)	Surgery (Y/N)	Surgical Details	HA Outcome, FU
Kewlani et al., 2020*	32, F	HA, neck pain, back pain, pressure behind the orbits	T2-3 osteophyte	2, 1	Y	Posterior laminectomy (T2-3), transdural discectomy, 1° repair of the ventral dural defect using 2 × interrupted 7-0 synthetic sutures, 2° repair using a ventral sling of bovine pericardium, lumbar drain placement	Resolved, 8 mos

B = blind; disc = herniated disc; FU = follow-up; HA = headache; ND = no data; T = targeted.

* Present study.

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Disclosures

Dr. Robbins reported serving on the board of directors of the American Headache Society and the New York State Neurological Society in nonremunerative positions. He receives an editorial stipend from Springer Publishing and book royalties from Wiley. No other disclosures were reported.

Author Contributions

Conception and design: Kewlani, Robbins. Acquisition of data: Greenfield, Kewlani, Garton, Hussain, Chazen. Analysis and

interpretation of data: Kewlani, Garton, Hussain, Chazen, Robbins, Baaj. Drafting the article: Greenfield, Kewlani, Garton, Hussain. Critically revising the article: Greenfield, Kewlani, Garton, Hussain, Chazen, Robbins. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Greenfield. Statistical analysis: Kewlani.

Supplemental Information

Video

Video 1. <https://vimeo.com/662928237>.

Previous Presentations

Portions of this work were presented as a poster at the ASIT (Association of Surgeons in Training) MedAll Virtual Summit Conference, October 27, 2020, and as a poster at the Global Spine Congress, November 3–6, 2021, virtual meeting.

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