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.5,6 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 hypertension; 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 0.3 × 0.2 × 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.

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

**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.
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...
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. 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 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. 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. 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, engagement of the venous structures, puititary hyperemia, and downward displacement of the cerebellar tonsils. 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. 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.

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). 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. 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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| 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 | 32, M | HA | C5–6 osteophyte | 1, 0 | Y | Anterior discectomy (C5–6), Osteophytectomy, 1° dural repair (2× interrupted Prolene sutures) | Resolved, ND |
| Eross et al., 2002 | 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 | 39, F | HA, neck stiffness, nausea, vomiting, sensitivity to changes in position | C7–T1 osteophyte + disc | 3, 0 | Y | Anterior cervical discectomy/osteophytectomy, fusion at C7–T1 with an allograft & metal plate, 1° dural repair, lumbar drain placement | Improved, 1 mos |
| Eross et al., 2002 | 46, F | HA, posterior neck pain, tinnitus, epistaxis | C4–5 osteophyte + disc | 1, 0 | N | | Improved, 2 mos |
| Winter et al., 2002 | 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 |
| Rapport et al., 2003 | 37, F | HA, interscapular pain, nausea, photophobia | T7–8 disc | 0, 2 | N | | Improved, ND |
| Binder et al., 2005 | 55, F | HA, aural fullness, memory difficulties, disorientation | T2–3 osteophyte | 1, 0 | Y | Anterior discectomy (T2–3), Osteophytectomy, 1° dural repair (2× nonabsorbable vertical mattress sutures & a small piece of interposed muscle --- fibrin glue) | Resolved, 12 mos |
| Yokota et al., 2008 | 25, F | HA, tinnitus | T8–9 osteophyte | 1, 1 | N | | Resolved, 24 mos |
| Witiw et al., 2012 | 46, F | HA, nausea, vomiting | C4–5 calcified disc | 3, 0 | Y | Anterior discectomy (C4–5), Osteophytectomy, 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 | 32, F | HA, neck pain & stiffness, nausea | T2–3 osteophyte + disc | 1, 0 | N | | Resolved, 4 mos |
| Hasiloglu et al., 2012 | 42, F | HA, dizziness, vertigo, nausea, vomiting | T5–6, T11–12 osteophytes + disc | 0, 1 | N | | Resolved, 6 mos |
| Allmendinger & Lee, 2013 | 56, F | HA | T6–7 herniated calcified disc | 1, 1 | N | | Resolved, ND |
| Allmendinger & Lee, 2013 | 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        | 49, M          | HA                       | T5–6 osteophyte + disc | 0, 2       | N             |                                                                                  | Resolved, ND   |
| Veeravagu et al., 2013          | 36, M          | HA, blurry vision, tinnitus | T1–2 osteophyte  | 1, 2       | Y             | Posterior hemilaminectomy (T1–2), partial corpectomy & facetectomy, osteophysectomy, DuraGen + morcellized Surgicel & fat graft (placed anteriorly), DuraGen sealed w/ Tisseel | Resolved, 36 mos|
| Wilson et al., 2013             | 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            | 57, M          | HA                       | T6–7 calcified disc | 0, 1       | N             |                                                                                  | Resolved, 5 wks |
| Rapoport et al., 2014           | 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                | 34, F          | Postural HAs, neck tightness | T2–3 osteophyte  | 2, 1       | N             |                                                                                  | Resolved, 20 mos|
| Pricola Fehnel & Borges, 2015   | 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               | 38, F          | HA, nausea, vomiting, muffling noise in the ears, worsening gait | T12–L1 osteophyte | Multiple, not specified | Y             | Hemilaminectomy (T12-L1), osteophysectomy, 1°, deemed not possible due to location of durotomy, DuraGen placed w/ addition of Tisseel, lumbar drain placement | Resolved, 4 yrs |
| Sartip et al., 2016             | 41, F          | Positional HAs, photophobia, nausea, vomiting | T5–6 calcified disc | 0, 2       | N             |                                                                                  | Resolved, 9 yrs |
| Urbach et al., 2019             | 34, M          | HA                       | T2–3 osteophyte  | 0, 0       | Y             |                                                                                  | Resolved, ND   |
| Fiechter et al., 2019           | 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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| Age (yrs) | Sex | Spinal Disorder | Symptoms at Presentation | Surgical Details | EBP | Surgery (Y/N) | Spine | Outcome | FU | Authors & Year |
|----------|-----|-----------------|--------------------------|-------------------|-----|---------------|-------|---------|----|----------------|
|          |      |                 |                          |                   |     |               |       |         |    |                |
| 32       | F    | 12–3 osteophyte | HA, neck pain, back pain, pressure behind the orbits | Resected laminectomy (T2–3), transdural discectomy, 1° repair of the ventral dural defect using 2/0 interrupted 7-0 synthetic sutures, 2° repair using a ventral sling of bovine pericardium, lumbar drain placement | 2/1 | Y             |       | Resolved | 8 mos | Kewlani et al., 2020* |
|          |      |                 |                          |                   |     |               |       |         |    |                |
|          |      |                 |                          |                   |     |               |       |         |    |                |
| 32       | F    | 12–3 osteophyte | HA, neck pain, back pain, pressure behind the orbits | Resected laminectomy (T2–3), transdural discectomy, 1° repair of the ventral dural defect using 2/0 interrupted 7-0 synthetic sutures, 2° repair using a ventral sling of bovine pericardium, lumbar drain placement | 2/1 | Y             |       | Resolved | 8 mos | Kewlani et al., 2020* |
|          |      |                 |                          |                   |     |               |       |         |    |                |
|          |      |                 |                          |                   |     |               |       |         |    |                |

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

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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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