Ossification of the ligament flavum (OLF) is a well-recognized cause of thoracic cord compression in the Asian population; however, its incidence has not been clearly identified in the Western hemisphere. The disease predominantly affects the lower thoracic spine, and approximately half of the patients with this disorder can have ossification of the underlying dura mater as well.6,11,13 Given the rarity of thoracic OLF, its clinical presentation remains unclear and often causes diagnostic ambiguity. It has often been overlooked or misdiagnosed as a lumbar spinal disorder, even by spine surgeons.19 It can be an incidental finding on MRI in asymptomatic patients, or it can present with back pain, progressive myelopathy, or even acute paraplegia after minor trauma.5,18 While various presentations have been reported, OLF has not been discussed as a cause of spontaneous cerebrospinal fluid (CSF) leakage causing intracranial hypotension and low-pressure headache. We describe the clinical, radiological, and intraoperative findings of a patient with OLF presenting with spontaneous CSF leakage to elucidate the pathology and to inform the readers of this rare presentation.

Case Report

History and Examination

A 50-year-old woman presented to the neurosurgery clinic with a 5-year history of progressive back pain. She did not have any radicular pain. She had undergone nonoperative treatment including physical therapy and oral pain medication, which only partially alleviated her pain. Additionally, over the prior 3 months, she had experienced progressively worsening postural headaches, worse while standing and relieved when lying down. Her neurological examination was normal. An MRI study of the brain, done for symptoms of spontaneous intracranial hypotension (SIH), was normal. Because of her continued back pain, MRI of the lumbosacral spine was performed
(Fig. 1), which demonstrated a stable L4–5 spondylolisthesis without central or foraminal stenosis. The rostralmost images of the same MRI study revealed a circumferential epidural fluid collection extending from T-10 to L-2 consistent with CSF. A CT scan revealed scalloping of the posterior margin of L1–3. A CT myelogram (Fig. 2) revealed OLF at T10–11. There was extravasation of intrathecal instilled contrast in the posterior epidural space at T10–11 on the right side. The ossified ligamentum flavum was suspected to be the cause of the leak. An epidural blood patch failed to be curative, and the patient was taken for operative repair.

Surgery

A T10–11 laminectomy was performed in a standard fashion, and when the epidural space was entered, a large amount of CSF under high pressure, consistent with a CSF leak, was encountered. The dura was inspected and, in addition to a pseudomeningocele capsule (arachnoid outpouching), there was an obvious violation of the dura in the axilla of the right T-10 nerve root due to erosion by the OLF. The OLF and medial portion of the hypertrophied facet joint were removed to allow further exposure of this region. There was chronically scarred arachnoid emanating from this defect in the dura on the right-hand side at the T-10 nerve root. The nerve root was suture ligated, the arachnoid outpouching was cauterized to reduce its size, and the dural defect was repaired using a running 6-0 Gore-Tex suture with a small piece of free muscle buttress (Fig. 3). A Valsalva maneuver was performed, and no further CSF leakage could be identified, indicating a watertight closure. Fibrin glue was applied to the repair, and the wound was then closed in layers.

Postoperative Course

The patient was discharged on the 3rd postoperative day with minimal incisional back pain and complete resolution of her headaches. At 6 weeks after surgery, she remained...
asymptomatic and MRI demonstrated resolution of the epidural CSF collection (Fig. 4). At the 6-month follow-up, she remained asymptomatic with MRI showing sustained resolution of the epidural CSF collection (Fig. 5).

Discussion

Spontaneous Intracranial Hypotension

Spontaneous intracranial hypotension is a condition of low CSF volume and pressure due to persistent CSF leakage through a dural defect along the neuraxis. This can lead to downward traction on the brain, causing headaches and possible acute neurological deterioration as a result of brain herniation. When patients present with low-back pain and symptoms of SIH, there is a diagnostic dilemma, especially as in our case in which MRI of the brain revealed no radiological signs of SIH. While one or more of the typical features, such as pachymeningeal enhancement, subdural fluid collections, engorgement of venous structures, pituitary hyperemia, and tonsillar herniation,

FIG. 3. Intraoperative photograph (A) obtained after completion of the T10–11 laminectomy and right facetectomy, showing arachnoid outpouching (arrow) through the dura from which CSF was leaking. Isolation of the right T-10 nerve root (arrow, B) after decompressing the pseudomeningocele. Ligation of the nerve root to better delineate the dural defect (arrow, C). Final suturing (D) of the dural defect caused by the ossified ligamentum flavum. Figure is available in color online only.

FIG. 4. Six-week postoperative axial (A) and sagittal (B) T2-weighted MR images of the thoracolumbar spine showing a T10–11 laminectomy defect with the usual postoperative changes and complete resolution of the epidural CSF collection around T11–12. Figure is available in color online only.
are present in a majority of patients, around 20% have a normal brain MRI study. Davidson et al. proposed an algorithm for the management of such cases. In the absence of intracranial findings, it is prudent to obtain an MR image of the spine with a CT myelogram to identify a possible site of leakage.

Our spinal imaging findings were suggestive of a leak at T10–11 with dense extravasation of contrast at that level. An initial blood patch prior to localizing the CSF leak has resulted in success rates ranging from 36% to 90%, whereas targeted patches for known leaks have resulted in clinical improvement in 87% of cases. In our patient, an epidural blood patch failed to provide improvement; therefore, surgery was planned as it was believed that the OLF, which had caused dural erosion, was the cause of the leak.

Ossification of the Ligamentum Flavum

The pathogenesis of OLF remains unclear. Many studies have described the possible contributions of mechanical, metabolic, and cell biological factors to the development and progression of OLF, but the involved mechanism remains poorly understood. A recent study established a relationship between bone morphogenic protein–2 (BMP-2) and OLF. Asymptomatic OLF is relatively common in the elderly Japanese population, with an estimated incidence of 4%–6%. It presents mainly in the 5th and 6th decades of life and when symptomatic is usually progressive and requires surgery.

Ossification of the ligamentum flavum is located below T-8 in 70% of cases. Common presentations include chronic back pain, posterior cord syndrome, and even myelopathy. Rarely, patients present with acute, unexpected spinal cord injury following an accident. In a large series of 51 Korean patients, the main symptoms included motor dysfunction (80%), sensory deficits (67%), and pain, numbness, and claudication (59%) in the lower extremities. Atypical symptoms included a burning sensation in the feet and a band-like pain in the chest or abdomen either with or without neurological deficits. In advanced stages, the incidence of neurogenic bladder and bowel dysfunction is also relatively high. However, OLF presenting as SIH has not been previously described. According to some reports, the duration of symptoms and preoperative neurological status tend to determine the postoperative outcome.

Given the high incidence of intraoperative durotomy and CSF leak during removal of the OLF, several surgical techniques have been described to enhance safe removal. Additionally, the incidence of acute neurological deterioration after surgical treatment of thoracic canal stenosis has been as high as 14.5%.

Some authors have reported that OLF tends to be most severe at the facet joints. In our case, the lateral portion of the OLF beneath the facet eroded the dura at the axilla of the exiting nerve root, resulting in a CSF leak that probably behaved similar to a ball-valve mechanism, causing a large epidural CSF collection with consequent intracranial hypotension and low-pressure headache. To repair the dural defect, resection of the medial portion of the facet and ligation of the nerve root were necessary. Fortunately, there was no underlying dural ossification.

Conclusions

Ossification of the ligamentum flavum is a rare radiological finding, and a clinical presentation of intracranial hypotension in symptomatic patients has not been described. The purpose of this brief report is to bring attention to a unique clinical presentation of thoracic OLF. Physicians and surgeons should consider this entity in cases of SIH when other etiologies are not evident.

References


Disclosures
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