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Thoracic ossification of the ligamentum flavum causing acute myelopathy in a patient with cervical ossification of the posterior longitudinal ligament: illustrative case

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BACKGROUND Ossification of the ligamentum flavum (OLF) has been well characterized as a distinct entity but also in tandem with ossification of the posterior longitudinal ligament (OPLL) in noncontiguous spinal regions. The majority of OLF cases are reported from East Asian countries where prevalent, but such cases are rarely reported in the North American population.

OBSERVATIONS The authors present a case of a Thai-Cambodian American who presented with symptomatic thoracic OLF in tandem with asymptomatic cervical OPLL. A "floating" thoracic laminectomy, resection of OLF, and partial dural ossification (DO) resection with circumferential release of ossified dura were performed. Radiographic dural reexpansion and spinal cord decompression occurred despite the immediate intraoperative appearance of persistent thecal sac compression from retained DO.

LESSONS Entire spinal axis imaging should be considered for patients with spinal ligamentous ossification disease, particularly in those of East Asian backgrounds. A floating laminectomy is one of several surgical approaches for OLF, but no consensus approach has been clearly established. High surgical complication rates are associated with thoracic OLF, most commonly dural tears/cerebrospinal fluid (CSF) leaks. DO commonly coexists with OLF, is recognizable on computed tomographic scans, and increases the risk of CSF leaks.

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KEYWORDS ossification of ligamentum flavum; OLF; ossification of posterior longitudinal ligament; OPLL; floating laminectomy; dural ossification

Ligamentous ossifications in the spine, such as ossification of the posterior longitudinal ligament (OPLL), are well-recognized causes of symptomatic spinal cord compression in the United States. Ossification of the ligamentum flavum (OLF), although exhaustively described in Asian populations over the past few decades, has rarely been reported in the North American population.[1–5]

Diffuse spinal ligamentous ossifications have been described with coexistence of OLF, OPLL, and other ligamentous ossifications throughout the spinal axis.[3,6–9] The coexistence of thoracic OLF within a larger subgroup of cervical OPLL patients has been reported by authors to be 30.8% (21 of 68 patients),[7] 44% (36 of 82 patients),[6] and as high as 64.6% (115 of 178 patients).[10] These so-called tandem ossifications, however, have not been commonly described in the North American population.[3] This case report describes an Asian American patient with cervical OPLL who became acutely myelopathic as a result of concurrent thoracic OLF.

Illustrative Case

A 42-year-old Thai-Cambodian American female presented with a 4-month history of severe low back, groin, and upper buttock pain radiating to the hips. She did not experience numbness or tingling of the lower extremities. She had greater left than right lower extremity weakness, causing gait and balance disturbance. Aside from chronic mild neck pain, she had no upper extremity pain,

ABBREVIATIONS BMP = bone morphogenetic protein; CSF = cerebrospinal fluid; CT = computed tomography; DO = dural ossification; JOA = Japanese Orthopedic Association; miR = microRNA; MRI = magnetic resonance imaging; OLF = ossification of the ligamentum flavum; OPLL = ossification of the posterior longitudinal ligament.

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sensory disturbance, weakness, or clumsiness of the upper extremities suggestive of significant, concurrent cervical cord compression. She reported urinary incontinence, diminished perineal sensation, and constipation but no bowel incontinence.

Her neurological examination in the upper extremities was notable for normal strength and sensation to pin/light touch. Her upper extremity reflexes were brisk with absent Hoffman's signs. Her lower extremity examination revealed increased tone, 3–4/5 weakness throughout, and decreased sensation to light touch. Her proprioception was normal. She had no Chaddock's, Bing's, or Babinski's reflexes or ankle clonus. She was ambulatory but with a paraparetic gait for which she used a walker.

Cervical spine magnetic resonance imaging (MRI) and computed tomography (CT) showed multilevel stenosis, OPLL, and compressive myelomalacia at C3–4 (Fig. 1). Lumbar spine MRI showed congenital/degenerative stenosis and moderately severe L4–5 and L5–S1 degenerative disc disease. Electromyography/nerve conduction velocity showed L5 and S1 myotome denervation.

Despite abnormal cervical spine imaging findings, the only clinical findings clearly referable to the cervical spine were neck pain and brisk upper extremity reflexes. The discordance between the diagnostic studies and physical examination suggested thoracic myelopathy as the main cause of her long tract signs and lower extremity weakness. Thoracic MRI and subsequent CT were performed, confirming severe cord compression at T10–11 consistent with ossification of the ligamentum flavum (Figs. 2 and 3). A distinct, focal ossified segment of dura was also identified (Fig. 3).

Urgent T10–11 laminectomy and resection of OLF were performed. As suggested by preoperative CT imaging, it was confirmed intraoperatively that the ligamentum flavum and underlying posterior dura were ossified. A floating laminectomy with OLF resection was performed, leaving a plaque of ossified dura that was circumferentially released from the adjacent dura. This was performed to potentially limit an anticipated large iatrogenic dural defect that would be difficult to repair. Nevertheless, a small rent of the arachnoid underlying the DO was encountered at the margin of the ossified dural plaque that was overlaid with DuraGen and DuraSeal (Integra LifeSciences).

FIG. 1. Cervical spine CT (**left**) and MRI (**right**). Extensive OPLL extending from C1–2 through C5–6, with markedly severe stenosis at C3–4. Compressive myelomalacia is denoted by the *arrow* on the MRI scan.

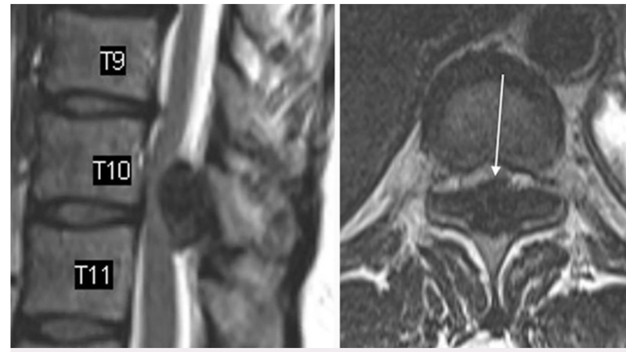

FIG. 2. Thoracic spine MRI. Sagittal view (**left**) showing severe T10–11 stenosis from a posterior calcified mass. Axial view (**right**) shows severe cord compression (*arrow*).

Despite normal neurophysiological recordings throughout the case, the patient awoke with dense worsening of her lower extremity paraparesis and severe postoperative back pain. She was discharged to inpatient acute rehabilitation on postoperative day 10. At her 15-month follow-up, her iliopsoas and quadriceps strength had improved to 5/5, and her overall lower extremity function had improved such that she no longer used a walker. She continued to have residual numbness and tingling of the right lower extremity, increased tone in the lower extremities, and dysesthetic pain around her thoracic incision site. Follow-up imaging showed satisfactory spinal cord decompression (Fig. 4). At 20 months after surgery, she developed a 1–2-month history of new, progressively worsening symptoms of severe proximal upper extremity pain, shoulder abductor weakness, and hand paresthesias. A multilevel laminoplasty was then performed for her acutely symptomatic cervical OPLL.

Discussion

The Sato classification of OLF describes 5 subtypes based on CT axial imaging: lateral, extended, enlarged, fused, and tuberous. A CT pictorial review and descriptions are detailed in Aizawa et al.,[11] Sun et al.,[12] and Miyakoshi et al.[13] The tuberous subtype, as in our case, is the most severe form, defined by complete transverse fusion of the ligamentum flavum with anterior extension. The tuberous subtype has also

FIG. 3. Thoracic spine CT. Sagittal view (**left**) shows OLF. Axial view (**right**) shows tuberous OLF subtype (*asterisk*) and associated "tram track" sign with distinct linear ossification consistent with DO (*arrow*).

FIG. 4. Immediate postoperative thoracic spine CT (**left**) shows residual plaque of "floating" DO (*arrow*). A 4-month postoperative thoracic spine MRI scan (**right**) shows dural tube reexpansion and decompression of T10–11. A postoperative pseudomeningocele without significant mass effect is noted.

been shown to be associated with a higher incidence of dural ossification (DO) and higher postsurgical cerebrospinal fluid (CSF) leakage.[12]

The prevalence of thoracic OLF is greatest in the lower thoracic spine.[6,11,14–21] In larger analyses, Kudo et al.,[20] in a study of 1,144 individuals, showed the most involved levels were at T9–L1, and Guo et al.,[21] in a study of 1,736 individuals, identified T9–10 to T10–11 as the more commonly affected levels. Liang et al.,[6] in a study of 2,000 individuals, found the highest incidence at T10 (24.1%), followed by T9 (14.2%) and T11 (11.9%). Lang et al.,[17] in a survey of 993 cases, showed 44% and 41.6% of OLF cases occurring at T10–11 and T11–12, respectively.

The higher incidence of OLF at T10–11 was speculated, by Maigne et al.[15] in a study of 121 cadaveric spines, to be due to increased rotational strain caused by the subjacent T11–12 level. A more rotationally restricted "lumbar" or sagittal facet orientation rather than a less rotationally restricted "thoracic" or coronal facet configuration at the T11–12 level was believed to cause more rotational stress of the T10–11 coronally oriented joints.

At the molecular genetic level, the involvement of signal transduction pathways in OLF-derived cells leading to osteogenesis has been described in several reports. The β-catenin,[22] Erk1/2, p38,[23] Indian hedgehog,[24] and Notch[25] signaling pathways appear to play a role in OLF osteoblast differentiation involving upregulation of osteogenic transcription factors. Expression of osteogenic growth factors, including cartilage-derived morphogenetic protein 1,[26] transforming growth factor-β, vascular endothelial growth factor, and bone morphogenetic proteins (BMPs) 2, 4, and 7, have also been identified in OLF specimens.[8,9,26]

Recent emerging studies on the role of microRNAs (miRs)—noncoding short segment nucleotides that regulate gene expression—have also recently been described in OLF osteogenesis.[27–29] Regulation of miR-targeted genes involved in OLF is an intriguing, futuristic therapy, but the only current effective treatment is surgical decompression.

Several surgical approaches for OLF have been reported, including thoracic laminectomy,[13,30,31] laminectomy with or without dural resection, duraplasty or dural "slitting,"[16,32] laminectomy with fusion,[30,31,33,34] laminoplasty,[33–35] en bloc laminectomy with or without ossified dural resection/duraplasty,[8,12,19,34,36] microendoscopic decompression,[37,38] and "floating" laminectomy.[8,13,32] Not unexpectedly, a consensus on the optimal surgical treatment for OLF remains ill defined.

The association of OLF and concurrent DO in the thoracic spine has been described with a prevalence of 13%–43.4%.[12,16,39,40] Muthukumar described the radiological "tram track" and "comma" signs as indicators of DO.[40] Recognition of DO in the setting of OLF is essential because this is associated with a high incidence of dural tears and CSF leaks. This can be as high as 93% compared with 9% in OLF patients without DO.[12] It is speculated that DO may be a local response to BMPs 2, 4, and 7 (osteogenic protein-1) identified in OLF specimens by immunohistochemistry.[9]

On the basis of the finding of DO associated with OLF in this case, a thoracic "floating" laminectomy and maximal safe OLF resection were planned. This included a circumferential release of the ossified dural plaque from surrounding normal dura to help facilitate dural tube expansion (i.e., "floating" of the DO and OLF remnant). Despite the intraoperative appearance of the dural tube not immediately reexpanding, postoperative imaging showed satisfactory reexpansion of the dural tube and cord decompression 4 months after surgery.

Complications of OLF surgery have been summarized in a systematic review and meta-analysis by Osman et al.[41] The pooled incidence rates showed a perioperative complication rate of 35%. Dural tears were the most common complication (18.4%), followed by CSF leak (12.1%), infection (5.8%), and neurological deficits (5.7%). It is unclear what distinguished a dural tear from a CSF leak in the reviewed literature, but it is separately reported here for historical accuracy. The meta-analysis by Osman et al. also demonstrated that despite some improvement in Japanese Orthopedic Association (JOA) scores, postoperative functional status remained poor after laminectomy alone.

Another systematic review of complications[42] after thoracic OLF surgery showed that the highest mean complication rate of 19% was due to CSF leaks, followed by a 5% mean complication rate due to neurological deterioration. The mean recovery rate based on pre- and postoperative JOA scores ranged from 31% to 68%. Complications of dural tears, as opposed to CSF leaks, were excluded due to the ambiguous distinctions as gleaned from their literature review.

In this report, we describe an Asian American female with tandem spinal ligamentous ossifications becoming acutely myelopathic as a result of thoracic tuberous subtype OLF. The pros and cons of a maximal OLF and DO resection and associated risk of a problematic CSF leak versus a somewhat less aggressive floating technique was contemplated preoperatively. A floating technique was selected to ideally minimize CSF leakage, with eventual pulsatile reexpansion of the dural tube indirectly assisting with cord decompression. This method assumes intradural pressures are sufficient to overcome opposing pressure from a developing epidural pseudomeningocele and/or postoperative seroma. This may not be as effective in cases of a larger persistent CSF leak and larger epidural fluid collections. This case shows that, with a smaller pseudomeningocele and relatively small arachnoid tear adjacent to retained but circumferentially released DO, dural reexpansion and cord decompression are possible. This occurred despite the immediate intraoperative observation of the dura appearing mechanically compressed anteriorly from residual posterior DO.

Although she had a satisfactory recovery, our patient eventually required a multilevel cervical laminoplasty 20 months later for subsequently symptomatic cervical OPLL. Thus, the complexity of caring for patients with OLF disorders is further illustrated.

The high incidence of spinal ligamentous ossifications in the East Asian population is well known. Consequently, the vast majority of the OLF literature and research has been reported from China, Japan, and Korea. In 2019, the U.S. population was estimated at 324,697,795, of which Asians represented 5.5%, or approximately 17,924,209 people.[43] Therefore, awareness of thoracic OLF as a cause of myelopathy, particularly in U.S. patients with East Asian racial backgrounds, should be considered in the differential diagnosis of myelopathy.

Observations

Thoracic OLF can coexist with asymptomatic cervical OPLL. Entire spinal axis imaging should be considered in the diagnostic work-up of myelopathy, particularly in individuals of East Asian backgrounds. A "floating" thoracic laminectomy with circumferential release of ossified dura and tuberous subtype OLF remnants can satisfactorily decompress the spinal cord in carefully selected patients. This may potentially limit larger iatrogenic and potentially problematic dural tears/CSF leaks. The stated observations are not unique in the Asian literature, but they have rarely been reported in the U.S. population. MRI illustrations of cord decompression after floating laminectomies of tuberous subtype OLF have not been commonly reported, to our knowledge. Awareness of tandem ossifications, and specifically OLF, is important for the North American neurosurgical community, based on current population demographic trends.

Lessons

High surgical complication rates are associated with thoracic OLF, with the most common being dural tears and CSF leaks, especially in the tuberous subtype. DO commonly coexists with thoracic OLF, can be recognized on preoperative imaging, and is associated with a high risk of dural tears/CSF leaks. The limitation of this report is that it describes a single patient treated with a floating laminectomy. Multiple surgical approaches are reported for the treatment of OLF, without a clear consensus on the optimal technique. Thus, selection of a treatment approach must be individualized for any given patient.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: both authors. Acquisition of data: Uchiyama. Analysis and interpretation of data: both authors. Drafting the article: both authors. Critically revising the article: both authors. Reviewed submitted version of manuscript: both authors. Approved the final version of the manuscript on behalf of both authors: Uchiyama.

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