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Case Report

Compressive Thoracic Myelopathy Caused by Combined Ossification of the Posterior Longitudinal Ligament and Ossification of the Ligamentum Flavum: A Report of Four Cases and a Literature Review
由合併胸椎後縱韌帶骨化及黃韌帶骨化引致的胸椎壓迫性脊髓病變 四病例報告及文獻回顧

Tong Jennifer Wing-Sze*, Chan Andrew Pak-Ho, Cheung Ka-Kin

Department of Orthopaedics and Traumatology, Tuen Mun Hospital, New Territories, Hong Kong

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ABSTRACT

We describe here the rare condition of compressive thoracic myelopathy and its management in four patients with combined thoracic ossification of the posterior longitudinal ligament and ossification of the ligamentum flavum. One of the four patients underwent decompressive laminectomies only, whereas the other three patients had posterolateral fusion with posterior instrumentation (pedicle screws and rods system) in addition to decompressive laminectomies. All four patients developed transient complete paraplegia after surgery. The three patients who received decompressive laminectomies and posterior instrumentation with posterolateral fusion had improved sensation, motor, and sphincter functions compared with their preoperative neurological state. The patient who underwent laminectomy only showed no neurological gain after 7.5 years. A dural tear was noted in two patients and they recovered without complications after intraoperative repair. Posterior instrumentation with posterolateral fusion in addition to decompression laminectomies and excision of the ossification of the ligamentum flavum seems to have a better outcome than simple decompression laminectomies for this rare cause of compressive thoracic myelopathy.

中 文 摘 要

我們報告一種罕有病例及其治療果效，由合併胸椎後縱韌帶骨化及黃韌帶骨化引致的胸椎壓迫性脊髓病變。在四個病例中，一人只進行了胸椎椎板切除術，其餘三人進行了胸椎椎板切除術，後植入物內固定和後外側脊柱融合術。術後四人均出現暫時性下身癱瘓，三名患者(進行了胸椎椎板切除和後外側脊柱融合術) 的感覺，運動和括約肌功能較術前有改善；另一患者(只進行了胸椎椎板切除術)在七年半後仍沒有神經功能的進步。有兩病例發生硬膜囊撕裂，縫合後順利康復。，後植入物內固定和後外側脊柱融合術加上減壓胸椎椎板切除及黃韌帶骨化切除術，似乎對這種罕有的胸椎壓迫性脊髓病變，有更好的果效。

Introduction

Thoracic ossification of the posterior longitudinal ligament (OPLL) and ossification of the ligamentum flavum (OLF) develop insidiously over a long period of time. This condition can have devastating consequences and seriously compromise the thoracic spinal cord. The diagnosis of thoracic myelopathy is often difficult because the symptoms are very vague and similar to other lumbar disorders. Treatment therefore tends to be delayed.

Case reports

Between 2004 and 2009, four patients with compressive thoracic myelopathy caused by OPLL and OLF at the thoracic level underwent surgery in our hospital (Table 1).The diagnosis of thoracic myelopathy was established by neurological examination, computed tomography, and magnetic resonance imaging. The magnetic resonance imaging scan showed that the spinal cord was compressed by OPLL and OLF at the thoracic level in all four patients (Figures 1–3). The pre- and postoperative neurological condition of each patient was evaluated using the modified Japanese

* Corresponding author. *E-mail:* jentong0324@yahoo.com.hk.

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Table 1
Summary of clinical characteristics of patients

Patient no., age (yr)	Pathology and treatment	Preoperative (kyphotic) Cobb's angle	Postoperative (kyphotic) Cobb's angle	mJOA (preoperative/postoperative)	Recovery ratio (%)	Presence of dural tear	Follow-up time (yr)
1 (female), 49	OLF (T5–8) + OPLL (T5–8) T5–8 Posterior decompression T5–8 Excision of OLF T5–9 Laminectomy T5–9	T5–9, 26.25°	T5–9, 24°	5/5	0	Yes, repaired	7.5
2 (female), 51	OLF (T7–9) + OPLL (T2, T6–9) T7–9 Posterior decompression T7–9 Excision of OLF T7–9 Laminectomy T7–9 T7–9 Posterior instrumented fusion	T5–11, 31.2°	T5–11, 23.1°	0/8	72.73	No	5
3 (female), 54	OLF (T4–5) + OPLL (T4–6) T2–6 Posterior decompression T4–5 Excision of OLF T3–5 Laminectomy T1–7 Posterior instrumented fusion	T1–7, 26.25°	T1–7, 20.63°	4/10	85.71	No	3
4 (male), 63	OLF (T1–2) + OPLL (T1–2) T1–3 Posterior decompression T1–2 Excision of OLF T1–3 Laminectomy C7–T3 Posterior instrumented fusion	T1–3, 14°	T1–3, 5.8°	4/8	57.14	Yes, repaired	2.5

mJOA score = modified Japanese Orthopaedic Association score for thoracic myelopathy (from 0–11).

Orthopaedic Association (JOA) scoring system, an 11-point scale measuring truncal sensory function, lower limb sensory and motor function, and bladder function. (Table 2).

The recovery ratio was defined as: recovery ratio = [(postoperative score − preoperative score)/(11 − preoperative score)] × 100%. The outcomes were ranked as good (≥50%), fair (10–49%), unchanged (0–9%), and worse (<0%).

The mean number of thoracic spinal levels affected by OPLL and OLF was 3.5 and 4.25 levels, respectively. The mean extent of thoracic laminectomies included 3.25 laminae (range 2–5 laminae). There was a reduction in the mean kyphotic angle of 6° after surgery. Preoperatively, the mean modified JOA score was 3.25/11. At the last follow-up, the mean score rose to 7.75. The mean recovery rate was 53.90%. The results were classified as good in three patients and unchanged in one patient.

All four patients developed transient paraplegia after surgery. The three patients who had posterior instrumented fusion had sensory, motor, and sphincter functional improvement over their preoperative neurological state. One patient showed no neurological gain after 7.5 years.

No patient had additional anterior decompression surgery via a thoracotomy. A small dural tear occurred in two patients due to ossification of the dura matter. The dural tears were primarily repaired. All leakages healed without complications, without prolonged leakage of cerebrospinal fluid into the drain bottles and the development of pseudomeningocele.

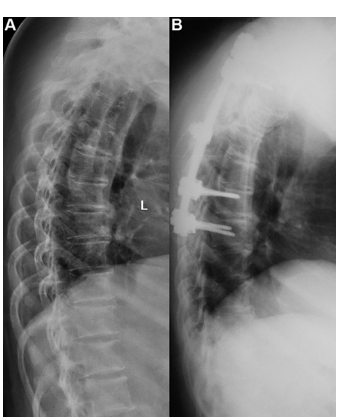

Figure 1. Patient 3. Ossification of ligamentum flavum T4–5 plus ossification of the posterior longitudinal ligament T4–6 with laminectomy T3–5 performed. (A) Preoperative and (B) postoperative radiographs of the thoracic spine.

Discussion

The prevalence of thoracic OPLL is 0.3–0.8% in the normal population[1] and it is more common in women.[2,3] Within the thoracic spine, OPLL is most commonly found in the upper and middle thoracic spine.[3] A large sample study[4] in China suggested that the overall prevalence of OLF within the southern Chinese population is 3.8%. Within the thoracic spine, OLF is commonly found in the lower and middle thoracic spine.

Combined thoracic OPLL and OLF can be asymptomatic, but are usually accompanied by myelopathy, with presentations of sensorimotor dysfunction in the trunk and lower extremities, and urinary disturbances.[5] The management of thoracic myelopathy due to combined OPLL and OLF is difficult. Conservative treatment is often ineffective and a gradual deterioration is seen.

Surgical intervention is also challenging both in terms of the surgical approaches and the complete removal of the thoracic OPLL and OLF. Sometimes it is technically impossible and the compromised spinal cord at the site of compression is vulnerable to damage during surgery. Dural tears are not uncommon. Furthermore, if the thoracic OLF posterior to the cord is also significantly compressing the spinal cord, anterior decompression will not be adequate even if the OPLL can be removed via a single anterior approach. Decompressive laminectomies with an excision of the

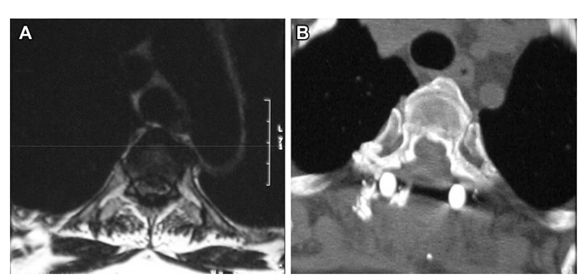

Figure 2. (A) Preoperative magnetic resonance imaging scan of T3–T4. (B) Postoperative computed tomography scan of T3–T4.

Figure 3. (A) Preoperative magnetic resonance imaging scan. (B) Postoperative magnetic resonance imaging scan.

thoracic OLF are not very effective due to natural kyphosis, which restricts a backward shift of the spinal cord.[6]

In a Chinese study,[7] 11 patients (8 men and 3 women) with thoracic myelopathy due to combined OLF and OPLL at the same thoracic level underwent posterior decompressive laminectomies and excision of the OLF. Posterior instrumentation and spinal fusion was performed in all patients. The thoracic kyphosis in the stabilization area was reduced from $30.0° \pm 4.02°$ to $20.8° \pm 2.14°$ on average. The mean modified JOA score improved from 3.5 ± 1.69 preoperatively to 8.5 ± 1.64 at the final follow-up, with a recovery rate of 68%.

The outcome in our series showed a lower recovery ratio (53.90%). This could be due to our small number of patients and the unsatisfactory result for our first patient with laminectomies only without instrumentation and fusion. For the three patients with additional instrumentation, an average recovery rate of 71.86% could be achieved, which is compatible with the Chinese study.[7]

All four patients developed transient paraplegia after surgery. In three patients, their lower limb muscle power dropped transiently by two to three grades. The lower limb motor power, sensation, and bladder function gradually improved with time. Only the first patient showed no neurological gain after surgery; she remained wheelchair-bound at 7.5 years follow-up. There was no improvement in her truncal and lower limb numbness. She had neurogenic bowel and bladder on self-intermittent catheterisation. Her modified JOA score remained unchanged.

The thoracic spine is naturally kyphotic and decompressive laminectomies are less effective because the backward shift of the spinal cord is restricted. The thoracic cord at the site of compression has a particularly vulnerable region called the watershed area with a poor blood supply. Iatrogenic catastrophic spinal cord injuries are more common here than with surgery in other parts of the spine, and this may explain the transient paraplegia.

Yamazaki et al[8] reported a case of recurrent thoracic myelopathy 4 weeks after anterior decompression and T3–T8 laminectomy using a posterior approach. The neurological deficits were corrected by a second operation with posterior instrumented T11–L1 fusion. They proposed that kyphosis and instability were the major factors potentially affecting the severity of thoracic myelopathy.

As a consequence of the unsatisfactory outcome of our first patient, we added additional posterior instrumentation and fusion with steroid cover (preoperative and postoperative dexamethasone, 2 mg four times per day for 3 weeks) for the subsequent patients. We also found that the subsequent patients had better kyphotic angle correction and stability postoperatively with instrumentation and fusion, which might contribute to better neurological recovery compared with the preoperative state. This echoes the proposal of Yamazaki et al.[8] We did not have any hardware complications of the instrumentation nor any postoperative infection. The mean follow-up period was 4.5 years (range: 2.5–7.5 years). The limitation of this study was the small sample size. From our review of the literature, only case series with different modalities of treatment methods and results could be found. Further control studies may be required to confirm the benefit of these techniques.

In conclusion, combined OPLL and OLF causing compressive thoracic myelopathy is a challenging clinical problem. Decompressive multiple level laminectomies, excision of the OLF, posterior instrumentation, and fusion can give a better stability of the spine and correction of the kyphotic angle after surgery, which may contribute to a better neurological outcome.

Table 2
Summary of modified Japanese Orthopaedic Association scores defining dysfunction due to thoracic myelopathy

Score	Description
Lower limb motor dysfunction	
0	Unable to walk
1	Able to walk on flat floor with walking aid
2	Able to walk up and/or down stairs with handrail
3	Lack of stability and smooth reciprocation of gait
4	No dysfunction
Lower limb sensory deficit	
0	Severe sensory loss or pain
1	Mild sensory loss
2	No deficit
Truncal sensory deficit	
0	Severe sensory loss or pain
1	Mild sensory loss
2	No deficit
Sphincter dysfunction	
0	Unable to void
1	Marked difficulty in micturition
2	Minor difficulty in micturition
3	No dysfunction

Conflicts of interest

The authors declare that they have no financial or non-financial conflicts of interest related to the subject matter or materials discussed in the manuscript.

References

1. Ohtsuka K, Terayama K, Yanagihara M, et al. An epidemiological survey on ossification of ligaments in the cervical and thoracic spine in individuals over 50 years of age. *J Jpn Orthop Assoc* 1986;**60**:1087–98.
2. Tsuyama N, Kurokawa T. Statistical analysis of the patients with ossification of the posterior longitudinal ligament of the thoracic and lumbar spines. *Rinsho Seikei Geka* 1977;**12**:337–9.
3. Yonenobu K, Ebara S, Fujiwara K, et al. Thoracic myelopathy secondary to ossification of the spinal ligament. *J Neurosurg* 1987;**66**:511–8.
4. Guo JJ, Luk KD, Karppinen J, et al. Prevalence, distribution, and morphology of ossification of the ligamentum flavum: a population study of one thousand seven hundred thirty-six magnetic resonance imaging scans. *Spine* 2010;**35**:51–6.
5. Jayakumar PN, Devi BI, Bhat DI, et al. Thoracic cord compression due to ossified hypertrophied ligamentum flavum. *Neurol India* 2002;**50**:286.
6. Yonenobu K, Sakou T, Ono K. *OPLL: ossification of the posterior longitudinal ligament*. Tokyo: Springer Verlag; 1997.
7. Zhang HQ, Chen LQ, Liu SH, et al. Posterior decompression with kyphosis correction for thoracic myelopathy due to ossification of the ligamentum flavum and ossification of the posterior longitudinal ligament at the same level. *J Neurosurg Spine* 2010;**13**:116–22.
8. Yamazaki M, Okawa A, Koda M, et al. Transient paraparesis after laminectomy for thoracic myelopathy due to ossification of the posterior longitudinal ligament: a case report. *Spine* 2005;**30**:E343–6.