

Progressive enlargement of thoracic ossification of the ligamentum flavum in professional baseball pitchers: a report of two cases

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Abstract

Both systemic and local factors might influence the development of ossification of the ligamentum flavum (OLF); however, whether specific physical activities play a role in the development of OLF remains unclear. This report presents two cases of thoracic radiculopathy owing to progressive enlargement of thoracic OLF within a few years in Japanese professional baseball pitchers. Two left-handed, Japanese, professional baseball pitchers in their 20s experienced thoracic radiating pain. Mild hypesthesia of the lower thoracic vertebral levels was observed, and an increase in the size of the OLF within a few years was identified in the lower thoracic spine using computed tomography and magnetic resonance imaging. Neither patient exhibited myelopathy and were, therefore, treated conservatively. Both patients were able to return to playing in top condition within 4 months after onset. The findings in these cases suggest that thoracic OLF might increase in size and become exacerbated in certain individuals, such as professional pitchers, who experience repeated, localized, mechanical stress on the thoracic spine, irrespective of age.

Keywords

Ossification, ligamentum flavum, myelopathy, radiculopathy, baseball, case report

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Introduction

Ossification of the ligamentum flavum (OLF) is widely recognized as a cause of thoracic myelopathy and radiculopathy in East Asia, and especially in Japan.^{1–8} OLF is a degenerative change in the spine that

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frequently occurs with aging, with most thoracic OLF patients being in their 50s or older; the pathology is also more frequent in men.⁹ Although the direct cause of OLF remains unclear, both systemic and local factors can influence its development. The most common spinal level in which OLF develops is the thoracic spine, and traction force over the course of many years under kyphotic spinal alignment is thought to influence ossification.^{10,11} The degree of OLF increased with age in a cross-sectional study, but the natural course contributing to OLF enlargement was unclear, although OLF is predicted to occur over time.⁷ Only four cases of symptomatic thoracic OLF in young athletes before their 30s have been reported,^{12,13} and whether specific physical activities (manual labor, sports) play a role in the development and enlargement of OLF remains unclear.

We report two cases of thoracic radiculopathy owing to progressive enlargement of thoracic OLF in Japanese professional baseball pitchers.

Case Report

The reporting of this study conforms to the CARE guidelines.¹⁴

Case 1

A 27-year-old, left-handed, Japanese professional baseball pitcher was referred to our institute because of intractable left chest and upper abdominal pain with numbness. He had no family history of ossification of the posterior longitudinal ligament or ligamentum flavum. Physical examination revealed radicular pain exacerbated by lateral bending, rotation, flexion, and extension of the trunk. The pain resolved during prolonged sitting and was completely relieved in the supine position.

Neurological examination revealed mild hypesthesia of the left T9–T11 levels without motor disturbance or abnormal tendon reflexes. Computed tomography (CT) revealed left unilateral or bilateral thoracic OLF at spinal levels T8/9, T9/10, T10/11, and T11/12, and magnetic resonance imaging (MRI) revealed foraminal stenosis without compression of the central cord and disc degeneration at segments T9/10 and T10/11 (Figure 1a–h).

Because no evidence of myelopathy was apparent, rest and physiotherapy were prescribed. The patient provided consent before receiving treatment. Physiotherapy comprised manual therapy, postural re-education, and exercise therapy aimed at minimizing thoracic kyphosis-induced spinal loading by increasing the strength and endurance of the back extensors.¹⁵ His symptoms gradually resolved, and he resumed physical exercise 2 weeks later. He was able to return to play in top condition 4 months after onset. He pitched professionally for 2 years before retiring, after which he threw an average of 150 pitches per day as a batting-practice pitcher for a professional baseball team from February to October each year. In his fifth year as a batting-practice pitcher, 7 years after injury onset, at the age of 34, the left chest pain recurred, and CT revealed enlargement of the OLF at all thoracic levels (Figure 1i–n). In particular, the OLF had fused bilaterally at the middle of the spinal canal at spinal levels T8/9 and T9/10 (Figure 1j, k). Conservative treatment proved ineffective this time, and he changed from work as a batting-practice pitcher to a scorer. His symptoms disappeared immediately after changing jobs, and he has continued working, without symptoms, for 2 years. Surgical decompression will be considered if he exhibits myelopathic symptoms; regular follow-up is on-going.

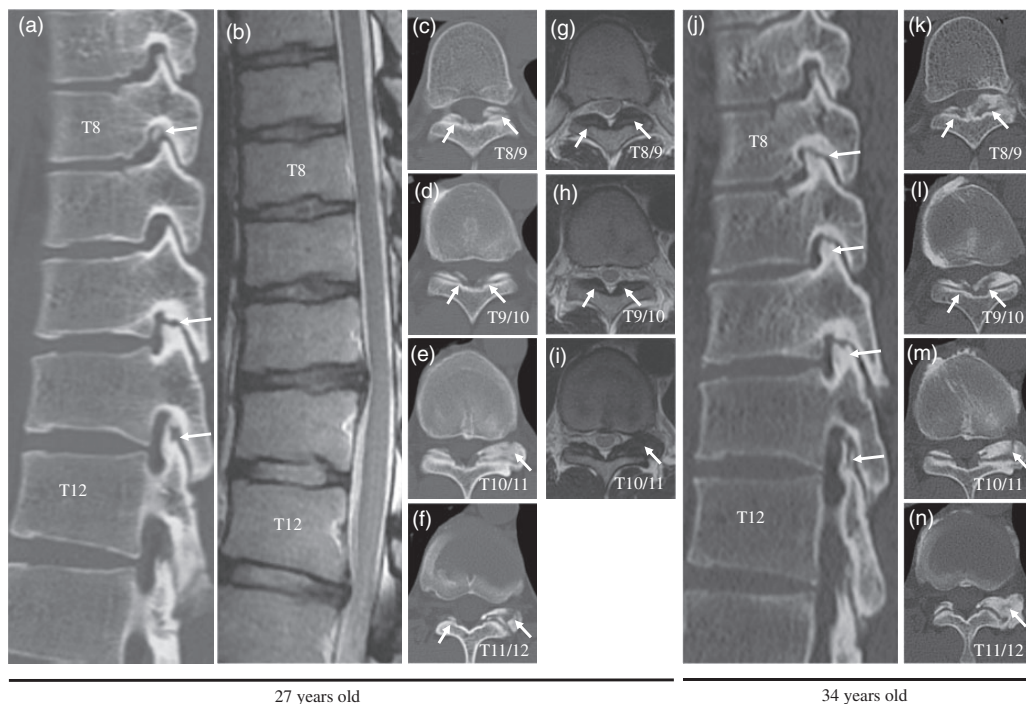


Figure 1. Case 1: Radiological findings in the thoracic spine on computed tomography and magnetic resonance imaging showing the presence of ossification of the ligamentum flavum (arrows). Left parasagittal view (a), sagittal view (b), and axial view at T8/9 (c, g), T9/10 (d, h), T10/11 (e, i), and T11/12 levels (f) of the spine when the patient was 27 years old. Left parasagittal view (j) and axial view at T8/9 (k), T9/10 (l), T10/11 (m), and T11/12 (n) of the spine when the patient was 34 years old.

Case 2

A 22-year-old, left-handed, Japanese, professional baseball pitcher was referred to our institute for a medical check-up. He had no family history of ossification of the posterior longitudinal ligament or ligamentum flavum. CT revealed mild, left-sided OLF at the T8/9 spinal level (Figure 2a, b), without neurological deficits. Two years after starting his professional career, he experienced chest wall pain in the left lower ribs. Neurological examination revealed hypesthesia of the left chest at the T8/9 spinal level. Thoracic CT showed enlargement of the OLF at the T8/9 level (Figure 2c, d), and MRI

showed no disc degeneration in all thoracic spinal levels (Figure 2e).

Rest and physiotherapy were prescribed because of the lack of myelopathy, and the patient provided consent before receiving treatment. He was able to gradually increase his level of physical exercise and returned to play in top condition 6 weeks after onset without recurrence of any symptoms.

Discussion

We presented two rare cases of thoracic radiculopathy owing to progressive expansion of OLF in professional baseball

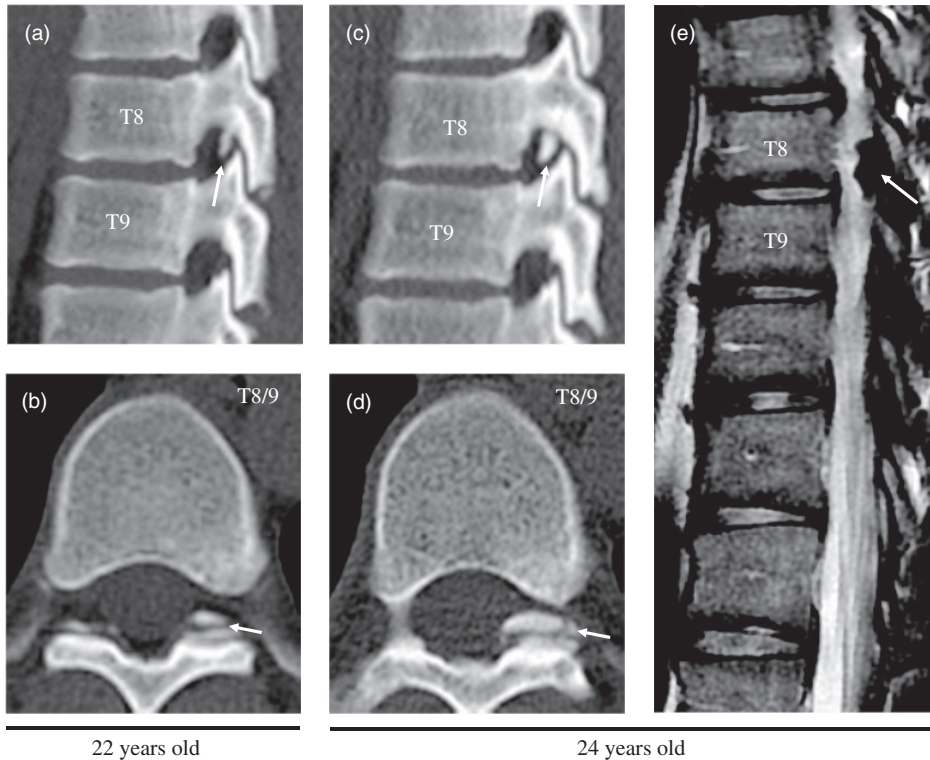


Figure 2. Case 2: Radiological findings in the thoracic spine on computed tomography and magnetic resonance imaging revealing the presence of ossification of the ligamentum flavum (arrows). Left parasagittal view (a) and axial view at T8/9 (b) when the patient was 22 years old. Left parasagittal view (c, e) and axial view of the spine at T8/9 (d) when the patient was 24 years old.

pitchers. Thoracic OLF might increase in size and become exacerbated among certain individuals, especially athletes, experiencing repeated, localized mechanical stress on the thoracic spine, irrespective of age.

Playing baseball, especially pitching, might be a risk factor for thoracic OLF enlargement. Several reports have described risk factors for OLF, namely ethnicity, genetics, metabolism, and systemic bone disorders.¹⁰ Although mechanical stress has been proposed as a possible cause of OLF,^{1,5,12,16,17} few clinical reports involving younger patients have indicated the critical role of repeated mechanical stress in developing ossification.^{12,13} The frequency and size of OLF were shown to depend on the

orientation of the zygapophyseal joints of the thoracic spine, with an increased range of rotation resulting in larger size and increased frequency.¹⁶ Repeated, localized, and asymmetrical rotatory mechanical stress caused by pitching motions may influence the development of OLF among high-level pitchers.^{12,13} In our cases, both patients became symptomatic in their 20s, and neither had the classical risk factors for OLF. Furthermore, the size of the OLF increased more rapidly than typical cases, which exhibit increasing size over longer periods of time.⁷ Both patients were highly active pitchers engaged in intensive training, including pitching almost every day. The baseball pitching motion involves a

distinct mechanism to facilitate high-velocity throws: the pitcher's trunk rotates rapidly, and large amounts of mechanical energy are transferred via extensive torque.¹⁸ In addition, professional pitchers perform this motion repeatedly during both games and training. In fact, eight Japanese professional baseball pitchers were reported in Japanese newspapers to have undergone surgery for thoracic OLF, whereas no affected fielders have been reported.¹⁹ According to our report and others, high-level East Asian baseball players, especially pitchers, might represent a high-risk population for symptomatic thoracic OLF. However, no reports have investigated the incidence of thoracic OLF in athletes. Therefore, further epidemiological study of thoracic OLF in athletes, including information regarding sports disciplines or specific positions, is needed.

Thoracic OLF rarely causes radiculopathy because ossification typically begins at the capsular portion and progresses to the interlaminar portion of the ligamentum flavum.²⁰ Most reported cases present with radiculopathy with myelopathy.^{5,21–23} In the present cases, particularly in Case 2, progressive expansion of ossification of the capsular portion led to foraminal stenosis and unilateral radicular pain without symptoms of myelopathy. Repeated traction force might unilaterally affect the ligamentum flavum owing to asymmetrical, rotational flexion-extension movements of the thoracic spine.^{12,15,16} We recommend that clinicians include thoracic spine pathology in the differential diagnosis during the diagnostic workup for chest wall pain in high-level baseball pitchers.²⁴

Both patients in this report were treated conservatively. Surgical treatment should be considered in patients with myelopathy and in those who do not respond to conservative therapy. Posterior decompression without fusion has been successful for ossified lesions in professional baseball

pitchers.^{12,13} Although neither patient in this study exhibited myelopathic symptoms, long-term follow-up is needed.

In conclusion, we presented two cases of thoracic OLF enlargement in Japanese professional baseball pitchers. These cases indicate that thoracic OLF might increase in size in certain individuals, including high-level baseball pitchers, experiencing repeated, localized mechanical stress on the thoracic spine. Further epidemiological study of thoracic OLF in baseball players is warranted.

Declaration of conflicting interest

The authors declare that there is no conflict of interest.

Ethics statement

At our institution, case reports do not require ethics review committee approval. The patients described in this paper provided written informed consent for publication.

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References

1. Yonenobu K, Ebara S, Fujiwara K, et al. Thoracic myelopathy secondary to ossification of the spinal ligament. *J Neurosurg* 1987; 66: 511–518.
2. Okada K, Oka S, Tohge K, et al. Thoracic myelopathy caused by ossification of the ligamentum flavum. Clinicopathologic study and surgical treatment. *Spine (Phila Pa 1976)* 1991; 16: 280–287.
3. Sato T, Kokubun S, Tanaka Y, et al. Thoracic myelopathy in the Japanese: epidemiological

- and clinical observations on the cases in Miyagi Prefecture. *Tohoku J Exp Med* 1998; 184: 1–11.
4. Shiokawa K, Hanakita J, Suwa H, et al. Clinical analysis and prognostic study of ossified ligamentum flavum of the thoracic spine. *J Neurosurg* 2001; 94: 221–216.
 5. Liao CC, Chen TY, Jung SM, et al. Surgical experience with symptomatic thoracic ossification of the ligamentum flavum. *J Neurosurg Spine* 2005; 2: 34–39.
 6. Aizawa T, Sato T, Sasaki H, et al. Thoracic myelopathy caused by ossification of the ligamentum flavum: clinical features and surgical results in the Japanese population. *J Neurosurg Spine* 2006; 5: 514–519.
 7. Mori K, Kasahara T, Mimura T, et al. Prevalence, distribution, and morphology of thoracic ossification of the yellow ligament in Japanese: results of CT-based cross-sectional study. *Spine (Phila Pa 1976)* 2013; 38: E1216–E1222.
 8. Ando K, Imagama S, Ito Z, et al. Predictive factors for a poor surgical outcome with thoracic ossification of the ligamentum flavum by multivariate analysis: a multicenter study. *Spine (Phila Pa 1976)* 2013; 38: E748–E754.
 9. Hirabayashi S. Ossification of the ligamentum flavum. *Spine Surg Relat Res* 2017; 1: 158–163.
 10. Otani K, Aihara T, Tanaka A, et al. Ossification of the ligamentum flavum of the thoracic spine in adult kyphosis. *Int Orthop* 1986; 10: 135–139.
 11. Yoshiiwa T, Miyazaki M, Kawano M, et al. Analysis of the relationship between hypertrophy of the ligamentum flavum and lumbar segmental motion with aging process. *Asian Spine J* 2016; 10: 528–535.
 12. Kaneyama S, Doita M, Nishida K, et al. Thoracic myelopathy due to ossification of the yellow ligament in young baseball pitchers. *J Spinal Disord Tech* 2008; 21: 68–71.
 13. Kato K, Yabuki S, Otani K, et al. Ossification of the ligamentum flavum in the thoracic spine mimicking sciatica in a young baseball pitcher: a case report. *Fukushima J Med Sci* 2021; 67: 33–37.
 14. Gagnier JJ, Kienle G, Altman DG, et al. The CARE guidelines: consensus-based clinical case reporting guideline development. *Headache* 2013; 53: 1541–1547.
 15. Briggs AM, Van Dieën JH, Wrigley TV, et al. Thoracic kyphosis affects spinal loads and trunk muscle force. *Phys Ther* 2007; 87: 595–607.
 16. Maigne JY, Ayrat X and Guérin-Surville H. Frequency and size of ossifications in the caudal attachments of the ligamentum flavum of the thoracic spine. Role of rotatory strains in their development. An anatomic study of 121 spines. *Surg Radiol Anat* 1992; 14: 119–124.
 17. Tsukamoto N, Maeda T, Miura H, et al. Repetitive tensile stress to rat caudal vertebrae inducing cartilage formation in the spinal ligaments: a possible role of mechanical stress in the development of ossification of the spinal ligaments. *J Neurosurg Spine* 2006; 5: 234–242.
 18. Kimura A, Yoshioka S, Omura L, et al. Mechanical properties of upper torso rotation from the viewpoint of energetics during baseball pitching. *Eur J Sport Sci* 2020; 20: 606–613.
 19. Kato K, Otoshi K and Konno S. Ossification of the ligamentum flavum in professional baseball players. *Kansetsugeka (Journal of Joint Surgery)* 2016; 35: 540–546. [in Japanese].
 20. Ahn DK, Lee S, Moon SH, et al. Ossification of the ligamentum flavum. *Asian Spine J* 2014; 8: 89–96.
 21. Miyasaka K, Kaneda K, Ito T, et al. Ossification of spinal ligaments causing thoracic radiculomyelopathy. *Radiology* 1982; 143: 463–468.
 22. Yabe Y, Honda M, Hagiwara Y, et al. Thoracic radiculopathy caused by ossification of the ligamentum flavum. *Ups J Med Sci* 2013; 118: 54–58.
 23. Kilic M and Kalali F. Intercostal neuralgia due to calcification of ligamentum flavum in DISH patients. *EC Orthopaedics* 2018; 10: 41–45.
 24. Kato K, Yabuki S, Otani K, et al. Unusual chest wall pain caused by thoracic disc herniation in a professional baseball pitcher. *Fukushima J Med Sci* 2016; 62: 64–67.