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# Symptomatic thoracic ossified ligamentum flavum in achondroplasia: A case report and literature review

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Case Report

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# ABSTRACT

Background: In a patient with achondroplasia, it is rare to encounter thoracic cord compression due to both spinal stenosis and ossification of the yellow ligament (OYL).

Case Description: A 33-year-old achondroplastic female presented with a progressive spastic paraparesis attributed to thoracic stenosis combined with T10-T11 OYL. Following a laminectomy, the patient demonstrated a marked recovery of neurological function.

Conclusion: This case and 12 others from the literature demonstrated that achondroplastic patients who present with myelopathy secondary to thoracic stenosis and OYL can be readily diagnosed with MR/CT and successfully treated utilizing decompressive laminectomies with/without fusions (i.e., for those crossing the thoracolumbar junction).

Keywords: Achondroplasia, Laminectomy, Lumbar canal stenosis, Ossification of the ligamentum flavum

# **INTRODUCTION**

Ossification of yellow ligament (OYL) in the thoracic spine has only rarely been reported to result in severe myelopathy in achondroplastic patients with thoracic stenosis.<sup>[2-5,10-12]</sup> Here, the authors present a 33-year-old myelopathic Iranian female with achondroplasia, thoracic stenosis, and T10-T11 OYL whose spastic paraparesis successfully resolved following a laminectomy alone.

# **CASE REPORT**

A 33-year-old achondroplastic female presented with a progressive spastic paraparesis/unsteady gait of 6 months' duration. In addition to the characteristic features of spinal achondroplasia on routine X-rays, the thoracolumbar MRI and CT studies confirmed the presence of moderate thoracic stenosis plus T10/T11 OYL (i.e., dorsolateral hypodense masses) [Figures 1-3]. Following a two-level thoracic laminectomy for decompression and excision of OYL/stenosis, the patient's myelopathy dramatically improved. The postoperative MR confirmed adequate decompression at the T10/T11 levels [Figures 4 and 5].

# DISCUSSION

Ossification of the ligamentum flavum (OLF) is well-known that involves patients from all over the world, including Iran (i.e., the US).<sup>[1,6-9]</sup> Although rare, these patients typically had

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underlying congenital thoracic stenosis making them uniquely susceptible to myelopathy due to the additional superimposition OYL.<sup>[2-5,10-12]</sup> Further, we were able to identify 12 similar thoracic cases of myelopathic achondroplastic patients with stenosis/OYL in the medical literature [Table 1]. Putting our case together with those 12 from the literature, there were nine males and four females averaging 49.1 years of age; eight were Japanese, two were East Indian, one was Thai, one Turkish, and one was Iranian. Spastic



**Figure 1:** Total spine radiographs. (a) Posteroanterior radiograph shows large pedicle cortex outline, note the interpedicular distance progressively decrease from thoracic to the lumbar spine. Mild coronal imbalance is also seen. (b) Lateral views demonstrate short, large pedicles, and lumbar canal stenosis along with mild sagittal imbalance.



**Figure 2:** (a and b) Thoracolumbar sagittal MRI; T1- and T2weighted images show a posteriorly located hypointense extradural mass compressing the spinal cord at T10-T11 level, compatible with ossified ligamentum flavum.

paraparesis was evident within 11 cases, while two had cauda equina syndromes (i.e., with thoracolumbar stenosis/



**Figure 3:** CT scan, axial views at T10-T11 level shows bilateral fused type ossification of the ligamentum flavum.



Figure 4: Photograph of the patient 6 months after surgery.



**Figure 5:** Postoperative thoracolumbar MRI, (a and b) T1 and T2 sagittal weighted images show appropriate decompression of lumbar and low thoracic spine.

Author	Year	Ethnicity/ sex	Age	Level	Preop deficits	Surgical management	Outcome
Takano <i>et al.</i>	1987	Japanese/M	36	T4-L5		Laminectomy	Poor
Takano <i>et al.</i>	1987	Japanese/M	58	T8-T11		Laminectomy	Good
Kataoka	1990	Japanese/M	52	T8-T11		Laminectomy	Poor
Nakahashi <i>et al</i> .	1991	Japanese/M	19	T4/5, T10-T12		Laminectomy	Good
Baba <i>et al</i> .	1992	Japanese/M	19	T4/5, T10-T12		Laminectomy	Good
Imamura <i>et al.</i>	1997	Japanese/F	19	T9-T12		Laminectomy	Good
Suzuki <i>et al.</i>	2008	Japanese/M	53	T9-T12		Laminectomy	Good
Saito <i>et al.</i>	2014	Japanese/M	75	L1-L4		Laminectomy	Good
Chakraborty et al.	2017	Indian/F	30	T10-T12		Laminectomy, instrumentation	Poor
Kachonkittisak et al.	2019	Thai/M	52	T10-L5		Laminectomy, instrumentation	Good
Gokcen and Ozturk	2019	Turkish/M	24	T10-L4		Laminectomy, instrumentation	Good
Nanda <i>et al</i> .	2021	Indian/F	34	T9-L5		Laminectomy, instrumentation	Good
Current case	2022	Iranian/F	33	T10-T11		Laminectomy	Good
Summary	1987-2022	Japan	Range	Nine thoracic		Nine laminectomy	Three
		Indian	19-75	Three		Four laminectomy +	poor 10
		Thai		thoracolumbar		instrumented fusion	good
		Turkish		One lumbar			0
D D 10		Iranian	26.1				
Preop: Preoperative, deficits: MM: Mild myelopathy, ModM: Moderate myelopathy, SM: Severe myelopathy							

Table 1: Summary of thoracic/thoracolumbar OYL with stenosis in achondroplastic adults with "PLEASE LOOK AT PREOP DEFICITS AND FILL IN "

OYL). MRI is the diagnostic study of choice for identifying achondroplasia accompanied by OYL, lumbar canal stenosis, and wedge vertebra, but CT studies best define OYL's exact shape, location, and extent of dural ossification.<sup>[2-5,10-12]</sup> The operations performed included nine thoracic laminectomies, one lumbar laminectomy, and three had thoracolumbar instrumented fusions [Table 1]. In all cases, meticulous detachment/excision of OYL required a microscope to avoid a cerebrospinal dural fistula and minimize trauma to the underlying cord/cauda equina.<sup>[1,6-9]</sup> Outcomes were good in 10 of 13 patients with minimal preoperative neurological deficits while with the three with more advance preoperative myelopathy exhibited only fair outcomes.

# CONCLUSION

OLF should be considered among the differential diagnoses for achondroplastic patients with thoracic spinal cord compression and OYL. Following diagnostic MR/CT studies, these patients typically do well with laminectomies alone, only rarely requiring fusions.

# Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

# **Conflicts of interest**

There are no conflicts of interest.

# REFERENCES

- 1. Epstein NE. Ossification of the yellow ligament and spondylosis and/or ossification of the posterior longitudinal ligament of the thoracic and lumbar spine. J Spinal Disord 1999;10:250-6.
- 2. Gokcen HB, Ozturk C. Ossification of the ligamentum flavum at the thoracic and lumbar region in an achondroplastic patient. World Neurosurg 2019;126:461-5.
- 3. Kachonkittisak K, Kunakornsawat S, Pluemvitayaporn T, Piyaskulkaew C, Pruttikul P, Kittithamvongs P. Congenital spinal canal stenosis with ossification of the ligamentum flavum in an achondroplastic patient: A case report and literature review. Asian J Neurosurg 2019;14:1231.
- 4. Nakahashi K, Baba H, Takahasi K, Kawahara N, Kikuchi Y, Tomita K. Achondroplasia with ossification of yellow ligament of the thoracic spine: Report of a case. Orthop Surg Traumatol 1991;34:397-400.
- 5. Nanda A, Bansal K, Gupta A, Verma K, Manghwani J, Chhabra HS. Multilevel thoracic and lumbar ligamentum flavum ossification in an achondroplasic-a rare presentation.

Spinal Cord Ser Cases 2021;7:69-9.

- Rahimizadeh A. Isolated proximal thoracic ossified ligamentum flavum causing paraparesis in a Caucasian: A case report and review of literature. World Spinal Column J 2012;3:38-43.
- Rahimizadeh A, Asgari N, Soufiani H, Rahimizadeh S. Ossification of the cervical ligamentum flavum and case report with myelopathy. Surg Neurol Int 2018;9:263.
- Rahimizadeh A, Habibollahzadeh P, Williamson WL, Soufiani H, Amirzadeh M, Rahimizadeh S. Symptomatic thoracic ossification of the ligamentum flavum in a patient with ankylosing spondylitis: Report of a case and review. Surg Neurol Int 2021;12:596.
- 9. Rahimizadeh A, Soufiani H, Amirzadeh M, Rahimizadeh S. Ossification of the ligamentum flavum of the lumbar spine in Caucasians: Case series. J Spine Neurosurg 2017;6:5.

- Saito K, Miyakoshi N, Hongo M, Kasukawa Y, Ishikawa Y, Shimada Y. Congenital lumbar spinal stenosis with ossification of the ligamentum flavum in achondroplasia: A case report. J Med Case Rep 2014;8:88.
- Sharma D. A rare case of ossification of ligamentum flavum presenting as dorsal myelopathy in achondroplasia. J Neurol Stroke 2017;7:254.
- Suzuki K, Kanamori M, Nobukiyo M. Ossification of the thoracic ligamentum flavum in an achondroplastic patient: A case report. J Orthop Surg (Hong Kong) 2008;16:392-5.

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