INTERNATIONAL JOURNAL OF CURRENT MEDICAL AND PHARMACEUTICAL RESEARCH
ISSN: 2395-6429, Impact Factor: 4.656
Available Online at www.journalcmpr.com
Volume 8; Issue 03(A); March 2022; Page No.119-122
DOI: http://dx.doi.org/10.24327/23956429.ijcmpr20220027

Research Article

OSSIFICATION OF LIGAMENTUM FLAVUM – AN UNUSUAL PRESENTATION: A CASE REPORT

Aditya Gowda., Shivam Bansal., Sudhakar and Bhaskar Sarkar

Dept of Orthopedics, AIIMS Rishikesh

ARTICLE INFO

Article History:
Received 06th December, 2021
Received in revised form 14th January, 2022
Accepted 23rd February, 2022
Published online 28th March, 2022

Key words:
Ossification of Ligamentum Flavum(OLF)

ABSTRACT

Introduction: In context of thoracic myelopathy, ossification of ligamentum flavum(OLF) has been described as one of the common associations. But the resolution of the presentation by just conservative management is quite unique. Such a scenario is a rare presentation with a few reported cases in the available literature.

Case Presentation: We present a case of post-traumatic thoracic myelopathy associated with ossification of ligamentum flavum in an adult patient who presented to us 5 days after injury. A thorough pre-op assessment was done. But in the due course, there was a rather significant recovery in the deficits in the due course of hospitalisation. Therefore the plan for his decompressive surgery was deferred and he was discharged. 3-month follow-up showed significant improvement in symptoms with near-normal power.

Conclusion: Thoracic myelopathy following Ossification of ligamentum flavum may pose a challenge for a clinician because of the associated difficulty in the treatment plan. Thorough work-up and planning play an anecdotal role in the management. And if there is improvement in the deficits and signs, it is advisable to wait and watch for the results.

INTRODUCTION

The ligamentum flava are broad paired spinal ligamentous structures that connect the adjacent laminae of the vertebral column extending from C-2 to S-1[1]. Ossification occurs as an acquired degenerative process through endochondral ossification of hypertrophied fibrous tissue within the ligaments. [2,3]. Such a disease presentation causing thoracic myelopathy is considered rare especially in young adults without any reasonable underlying pathology. Such a patient presentation undergoing spontaneous resolution of symptoms and signs is a rare entity and have not been documented in the available literature. Moreover, most cases of OLF require some or the other active surgical intervention to give satisfactory relief for the patient. We present a case of Idiopathic Ossified Ligamentum flavum in which it was diagnosed as the cause for his deficits, but later on, he was free of symptoms 3 months follow up without any surgery.

Case Report

History

A 29 years old Indian male, presented with complaints of numbness in the bilateral lower limb and inability to walk. He had a history of fall from the standing height 5 days prior to presentation. His laboratory investigations were within the normal limits, S. UREA: 28.7mg/dL, S. CREATININE: 0.85 mg/dL, Na:141.8 Mmol/L, K: 4.2 Mmol/L, SGOT: 30.9 U/L, SGPT: 30.1 U/L.

Examination

Vertebral inspection revealed a central furrow. No paraspinal fullness, obvious deformity or swelling was appreciated. There were no open wounds, scars or sinus present in the paravertebral region. Palpatory findings were inconclusive. No tenderness or local rise of temperature was found.

Neurological examination was done according to ASIA charting:

Bulk comparable in Bilateral lower limb. Tone increased in Bilateral lower limb.

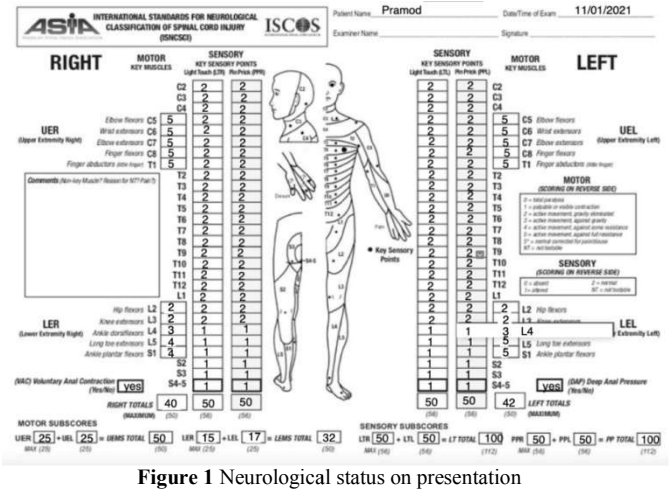

Figure 1 Neurological status on presentation

Corresponding author:* **Aditya Gowda
Dept of Orthopedics, AIIMS Rishikesh

Table 1 below incorporates the review of literature.

Author	Age/sex	Duration	Associated etiology	Deranged lab parameters	Neurology at presentation	Treatment given if any	Outcome
El Helou *et al* (2016)	33y/M	1yr	Idiopathic OLF Army officer-continuous mechanical stress on his spinal cord	Ligamentum flavum specimen was not sent for pathological laboratory study	Back pain with progressive worsening of gait Paraesthesia below level of nipple 2/5 motor strength in the lower limb bilaterally Hyperreflexia with Babinski sign	Laminectomy and flavectomy extending from T-2 to T-10 using high speed drill	Motor strength improved progressively over 6 months with intensive physiotherapy program in a specialized center. Paraesthesia resolved progressively, but hyper-reflexia persisted
Tokala *et al* (2007)	45y/M	3week		Haematological investigations and plain radiographs were normal	Acute progressive lower limb weakness, walking difficulties Loss of sensation below the epigastrium Bilateral ankle clonus, lower limb hyper reflexia with babinski sign	Bilateral laminectomies at T3, T4 and T5 f/b generous decompression of spinal cord	10 months follow-up, he was walking unaided within the house, was using one stick for short distances and a wheelchair for long distances. However, there was persistent bilateral lower limb hyper-reflexia, up-going plantars but no ankle cloni.
Tang *et al* (2017)	55/F	1yr	Multiple ossification of ligamentum flavum (OLF) T3 to L1 levels	The serum CRP, ESR,ANCA, igg levels were within the normal ranges. ANA and RF were negative	Weakness and numbness in B/L lower extremity 4/5 muscle strength	T3-L1 laminectomy	Five days post of surgery, the patient felt a reduced numbness and improvement in motor function.
Liu *et al* (2020)	70/M	1yr	Cervical stenosis caused by CLF (C5/C6) with myelopathy		Numbness in upper and lower extremities, disturbance of finger coordination, and gait disturbance A positive Babinski sign Power 4/5	Endoscopic decompressive surgery	Allowed to ambulate the next day after the operation and showed significant improvement of hand function and ambulation ability
Miura *et al* (2020)	78/M	1month	Idiopathic T10/11		Numbness and progressive gait difficulties Incomplete spastic paraplegia (3/5 power) Hyperactive deep tendon reflexes	Posterior decompression followed by instrumented fusion from T9–T12	He gradually recovered from the gait disturbance and could ambulate with the aid of a walker six months after
Miyakoshi *et al* (2019)	76/M	9months T11-T12 level	Osteoporotic vertebral fracture at the T11 and L1 H/O rx of the hypopharynx, early gastric cancer, and autoimmune hepatitis, with the latter treated using 6 mg/day of prednisolone for several years	Laboratory data showed no specific findings other than osteoporosis.	Impairment of standing resulting from progressive paraplegia showing weakness of the lower extremities (power 1-2/5)	Posterior decompression of T11-T12 with resection of the CLF and posterior instrumented fusion from T10 to L2 combined with vertebroplasty using hydroxyapatite blocks for the fractured T12 vertebra	Muscle weakness recovered completely, and the patient became able to walk with a cane.
Shiguematsu et al., 2012	47/F	T10-T11			Spastic paraparesis		
Sohail *et al* (2018)	43/M	Several days	Patient with hypoparathyroidism	Low ionized calcium level (1.99 mg/dl), low calcium level (4.2 mg/dl), low corrected calcium (5.0 mg/dl), low magnesium(0.9 mg/dl), high phosphorus (7.7 mg/dl), and a low parathyroid hormone level (<3.0 pg/ml)	Backache, lower extremity weakness, and urinary retention Severe spastic paraplegia; marked lower extremity weakness/paraparesis (1/5), bilateral diffuse hyperreflexia, and bilateral Babinski responses	Laminectomies at the C4-C5, T3-T4, and T10-T11 levels	The patient was discharged on the 10th postoperative day with a corrected calcium level of 7.8 mg/dl, having attained complete resolution of his preoperative motor deficit and urinary incontinence.
Chakravarthy *et al* (2014)	45/M	2yr D9 to D12	Undetected Hypoparathyroidism	Pre-operative investigations were normal. Serum procalcitonin 4.4 ng/ml, hypocalcaemia (serum calcium 4.1 mg%), hypoalbuminemia (2 g%)	Low backache and weakness of both lower limbs (1/5) and urinary retention and constipation last 2 days	Laminectomy and decompression of the spine.	
Martines *et al* (2012)	44/F	2months T6 and T10-11	Idiopathic endochondral calcification	Laboratory test results were normal, metabolic disorders being therefore ruled out.	Ascending paraesthesia of the left leg, with progressive worsening and right leg involvement, accompanied by reduced (right and left) leg muscle strength.	Laminectomy and dissection of the affected LF	After three months of clinical follow-up, the patient had progressed favourably, having no sensory complaints and again becoming ambulatory
Yang *et al* (2013)	26 patients	46.7 Months	Dural ossification in the thoracic spine		Paraparesis (24 patients), sensory change (25 pa- tients), tingling sensations (21 patients) and/or truncal or back pain (18 patients)	En bloc resection of the OLF and DO by cutting the bony junction between the pedicle and upper facet with a high-speed drill	The initial symptoms were significantly alleviated postoperatively. All 26 patients had postoperative CSF leakage,
Zhong *et al* (2016)	22 patients	35.6 months			Numbness and sensory dysfunction of the lower extremities in 18 patients (81.8%), weakness of the lower extremities in 13 patients (59.1%), gait disturbance in seven patients (31.8%), tightness sensa- tion of the trunk in three patients (13.6%), urinary disturbance in four patients (18.2%), and local back pain and/or leg pain in 11 patients (50.0%).	Posterior decompressive laminectomy was performed in all patients	Using a modified JOA score, 9 patients showed excellent results, 8 good, 4 fair, and 1 unchanged. The mean recovery rate was 65.49%.

Ho Li *et al* (2012)	31 cases			Symptoms of lower extremity numbness, lower extremity weakness, ambulatory diffi- culty/gait disturbance, severe thoracic myelopathy or radi- culopathy, urinary retention, and bladder and/or bowel incontinence 12-month history of pain in the thoracic spine and sensory disturbance of the lower extremities. Six months before admission he developed a progressive paraparesis.	1st group(13) underwent posterior decompression, 2nd group(7) underwent circumferential decompression, and the 3rd group(11) - posterior decompression and fusion group	Mean recovery rate at the final follow-up was found to be 46.5% in the 1st group, 65.1% in the 2nd group, and 62.7% in the 3rd group
Hepgul *et al*(1991)	59 yr/M	Paget's disease.	Histology showed cancellous bone with the typical appearances of Paget's disease.		Calcitonin was given	It is effective in reducing the excessive bone resorption of Paget's disease, in restoring biochem- ical parameters, and in the stabilization of radiological

The symptoms present did not correlate with the motor deficits. MRI lumbar spine was conclusive of pre/paravertebral collections causing compressive myelopathy with posterior element involvement.

He was diagnosed with Extradural compressive myelopathy secondary to intracanal callus from D9-D10.

The patient was planned for surgery, but the patient denied it. Hence he was managed conservatively. To our surprise, the patient improved dramatically. His ASIA scores improved within a week. The paraesthesia and tingling sensation also improved. The power in lower limbs started increasing.

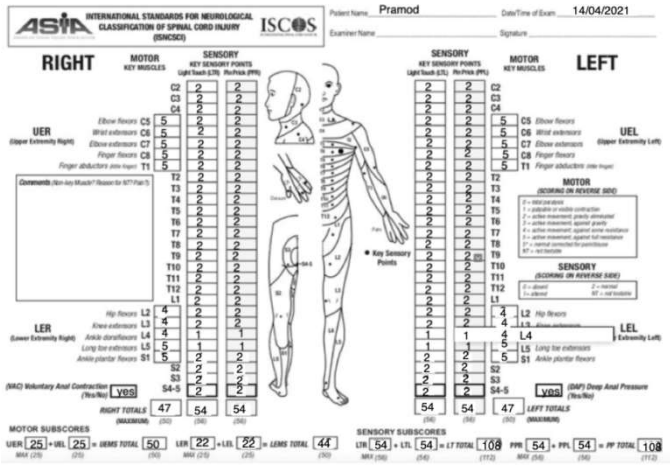

Figure 2 Neurological status at 3 months follow up

DISCUSSION

Ossification of the ligamentum flavum (OLF) is a well-recognized cause of myelopathy with a part of cord commonly affected in this condition. In the Asian region, especially in the Japanese race, the prevalence of OLF especially in the Thoracic region causing compression and neurological signs are frequently seen[4]. Whether OLF is more common in the Japanese population or is there anyracial difference is still unclear as most of the literature available on OLF is based on the prevalence of the condition in the Japanese population.

The pathogenesis of OLF remains unclear. Few case reports have reported its association with Parathormone disorders[5], diffuse idiopathic skeletal hyperostosis (DISH)[6], genetic causes (Runt-related transcription factor 2s)[3], skeletal fluorosis, Paget's disease[7], X-linked hypophosphatemia[8]. Many studies have described the possible contributions of mechanical, metabolic, and cell biological factors in the development and progression of OLF, but the involved mechanism remains poorly understood[9]. A recent study established a relationship between bone morphogenic protein–2 (BMP-2) and OLF[10].

In 51 Korean patients, the main symptoms present were motor dysfunction (80%), sensory deficits (67%), pain, numbness, and claudication (59%) in the lower extremities[11]. Atypical symptoms such as burning sensation in the feet and a band-like pain in the abdomen or chest region either with or without neurological deficits. In advanced stages, the incidence of neurogenic bladder and bowel dysfunction is also relatively high [12].

There are instances where conservative management has been tried for symptoms such as back pain and numbness and pain in the lower extremities, nonsteroidal anti-inflammatory drugs, muscle relaxants, use of vitamin B12 combined with outer compress and cream. Palliative drugs are additionally administered for severe neuralgic pain in the lower extremities.

Hepgul *et al*(1991) have described the use of calcitonin in cases of spinal Pagets. Calcitonin has shown improvement in neurological functions and has been able to reduce bone resorption and restore biochemical parameters. Though they have described the surgical treatment to be superior to calcitonin. [7]

Decompression surgery is recommended for patients with failure of conservative treatment & patients with severe spastic gait, severe muscle weakness of the lower limbs, and bladder-bowel involvement. Early surgery is indicated in patients presenting with OLF and OPLL at the same level as spinal cord is severely compressed both anteriorly and posteriorly. The surgical methods described are open-door type laminectomy, en bloc laminectomy, fenestration, and hemilaminectomy.

Yang *et al* (2013) in their clinical prospective study on 26 patients, described the outcomes of their decompressive surgery and followed them up 22–66 months (mean, 46.7 months) duration. the neurological status had improved at follow-up from a preoperative mean of 5.46 ± 1.73 (range, 2–9) points to 8.92 ± 1.38 (range, 5–11) points ($\mathbf{t} = 13.87$, $\mathbf{p} < 0.05$). The outcome of the intervention was found to be exceptional in six (23.1%) patients, good in 18 (69.2%) patients, and fair in two (7.7%) patients. No patient developed worsening of neurological symptoms. [13]

Zhong *et al* (2016) reported the results of 22 patients who underwent posterior decompressive laminectomy. An evaluation was done using a modified JOA score, 9 patients showed excellent results, 8 good, 4 fair, and 1 unchanged. The mean recovery rate was 65.49%. However, surgical complications occurred such as a Dural tear in five patients and CSF leakage, immediate postoperative neurologic deterioration, epidural hematoma, and wound infection in one patient each, respectively. [14] Mo Li *et al*(2012) reported results of 31 patients of OLF. Patients were divided into 3

groups based on surgery performed- 1st group(13) underwent posterior decompression, 2nd group(7) underwent circumferential decompression, and the 3rd group(11) - posterior decompression and fusion group (laminectomy was done along with posterior instrumented fusion). The mean recovery rate at the final follow-up was found to be 46.5% in the 1st group, 65.1% in the 2nd group, and 62.7% in the 3rd group. [15]

Our patient presented early with symptoms of 5 days duration following (fall from standing height) with complaints of inability to walk or stand up, which is an atypical presentation of complaints in the presence of motor deficit that doesn't correlate to the amount of symptom clinically. He also complains of numbness and tingling sensation in bilateral lower limbs following the episode.

In combination, they form the most sensitive and specific modality of investigation for the diagnosis of ossified ligamentum flavum. It is so because of the sensitivity of MRI to tissue hydration which helps in early diagnosis compared to other modalities of investigation. And CT is sensitive in picking up the ectopic location of calcification. In our case, the initial neurological deficiency was documented, serial daily ASIA charting was done to monitor and assess the neurology and the patient was managed conservatively while being prepared for surgical intervention if required.

In a span of 2 weeks, we witnessed a spontaneous recovery in his neurological status without any intervention done surgically. And this was supported even by radiology wherein there was spontaneous resolution of amount of calcification in the spinal canal compressing onto the spinal cord. Indications for surgical intervention and their outcomes in such similar patients are discussed in the table given below (review of literature). Our patient did not require any surgical intervention and to the best of our knowledge in English literature, spontaneous resolution of OLF has rarely occurred clinically and radiologically without any adverse sequalae and the patient returning back to his normal daily activities.

CONCLUSION

Thoracic myelopathy following Ossification of ligamentum flavum may pose a challenge for a clinician because of the associated difficulty in the treatment plan. Thorough work-up and planning play an anecdotal role in the management. And if there is improvement in the deficits and signs, it is advisable to wait and watch for the results as spontaneous remission might alleviate the need for surgical intervention.

Clinical Message

Serial neurological evaluation is necessary for assessment and it is advisable to wait and watch in cases where slight improvement is seen. This might alleviate the need for surgery and would reduce morbidity in such patients.

Authors contributions

"AG & S evaluated the patient and analysed the data. AG & SB went through the review of literature and wrote the manuscript, AG being the major contributor. BS finalised the manuscript and approved the final manuscript."

References

1. Gray H. Anatomy of the Human Body. Lea &Febiger; 1878. 1026 p.
2. Christiano LD, Assina R, Goldstein IM. Ossification of the ligamentum flavum: a unique report of a Hispanic woman. Neurosurgical Focus. 2011 Mar 1;30(3):E15.
3. Hirabayashi S. Ossification of the ligamentum flavum. Spine SurgRelat Res. 2017 Nov 27;1(4):158–63.
4. Gao R, Yuan W, Yang L, Shi G, Jia L. Clinical features and surgical outcomes of patients with thoracic myelopathy caused by multilevel ossification of the ligamentum flavum. The Spine Journal. 2013 Sep 1;13(9):1032–8.
5. Abdelmonem H. Ossified ligamentum flavum causing compressive myelopathy | Radiology Case | Radiopaedia.org [Internet]. Radiopaedia. [cited 2021 May 15]. Available from: https://radiopaedia.org/cases/ossified-ligamentum-flavum-causing-compressive-myelopathy
6. Guo Q, Ni B, Yang J, Zhu Z, Yang J. Simultaneous ossification of the posterior longitudinal ligament and ossification of the ligamentum flavum causing upper thoracic myelopathy in DISH: case report and literature review. Eur Spine J. 2011 Jul;20 Suppl 2:S195-201.
7. Hepgül K, Nicoll JAR, Coakham HB. Spinal cord compression due to pagetic spinal stenosis with involvement of extradural soft tissues: A case report. Surgical Neurology. 1991 Feb 1;35(2):143–6.
8. Untreated hypophosphatemic vitamin D-resistant rickets with symptomatic ossification of the ligamentum flavum. - Abstract - Europe PMC [Internet]. [cited 2021 May 16]. Available from: https://europepmc.org/article/med/1666964
9. Turel MK, Kerolus MG, O'Toole JE. Ossified ligamentum flavum of the thoracic spine presenting as spontaneous intracranial hypotension: case report. Journal of Neurosurgery: Spine. 2018 Apr 1;28(4):401–5.
10. Qu X, Chen Z, Fan D, Xiang S, Sun C, Zeng Y, *et al*. Two novel BMP-2 variants identified in patients with thoracic ossification of the ligamentum flavum. European Journal of Human Genetics. 2017 May;25(5):565–71.
11. Kang K-C, Lee C-S, Shin S-K, Park S-J, Chung C-H, Chung S-S. Ossification of the ligamentum flavum of the thoracic spine in the Korean population: Clinical article. Journal of Neurosurgery: Spine. 2011 Apr 1;14(4):513–9.
12. Jia L, Chen X, Zhou S, Shao J, Zhu W. En Bloc Resection of Lamina and Ossified Ligamentum Flavum in the Treatment of Thoracic Ossification of the Ligamentum Flavum. Neurosurgery. 2010 Jun 1;66(6):1181–6.
13. Yang Z, Xue Y, Zhang C, Dai Q, Zhou H. Surgical treatment of ossification of the ligamentum flavum associated with dural ossification in the thoracic spine. Journal of Clinical Neuroscience. 2013 Feb 1;20(2):212–6.
14. Zhong Z-M, Wu Q, Meng T-T, Zhu Y-J, Qu D-B, Wang J-X, *et al*. Clinical outcomes after decompressive laminectomy for symptomatic ossification of ligamentum flavum at the thoracic spine. J ClinNeurosci. 2016 Jun;28:77–81.
15. Li M, Meng H, Du J, Tao H, Luo Z, Wang Z. Management of thoracic myelopathy caused by ossification of the posterior longitudinal ligament combined with ossification of the ligamentum flavum-a retrospective study. Spine J. 2012 Dec;12(12):1093–102.