Ossification of the cervical ligamentum flavum and case report with myelopathy

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Abstract

Background: Ossification of the ligamentum flavum (OLF) occurs mostly in adult males, typically in the thoracolumbar spine where it may contribute to neurological deficits. Here we reviewed 68 cases of cervical OLF resulting in progressive quadriparesis.

Methods: The literature on cervical OLF was reviewed between 1962 and 2018 along with the case of an 81-year-old male with progressive quadriparesis attributed to cervical OLF.

Results: Most patients with cervical OLF are Asian, with Caucasians constituting the second most frequently impacted population.

Conclusions: Cervical OLF is typically reported in the Asian, followed by the Caucasian population, and is most often found in the thoracolumbar spine. Here we presented an 81-year-old male with cervical OLF contributing to quadriparesis.

Key Words: Cervical myelopathy, cervical spine, ossification of ligamentum flavum, ossification of posterior longitudinal ligament, ossification of yellow ligament, review article

INTRODUCTION

Ossification of the ligamentum flavum (OLF) typically occurs in adults involving the thoracolumbar spine. It is less frequently encountered in the cervical region (<1%). In 1962, Koizumi described cervical OLF at autopsy in a 55-year-old male who had developed progressive quadriparesis. Since then only 68 more cases of cervical OLF have been published, typically involving the East Asian populations presenting varying degrees of myelopathy.

Here, an 81-year-old Caucasian male with unilateral large nodular OLF at the C4-C5 level and a smaller lesion at C5-C6 presented with quadriparesis adequately treated with laminectomy and OLF resection.

CASE REPORT

An 81-year-old bed-ridden male presented a spastic quadriparesis. The T1-weighted sagittal magnetic resonance image (MRI) showed a large heterogeneous mass resulting...
in dorsolateral cord compression at C4-C5, and a smaller lesion at the C5-C6 levels, consistent with OLF [Figure 1]. On the T2-weighted MRI, the mass was isointense with a hypointense peripheral rim [Figure 1]. The CT scan confirmed ossification of both OYL lesions [Figure 2].

The patient underwent a C4-C6 laminectomy for resection of large dorsolateral OLF masses (C45, C56) [Figure 3]. Neurolysis and durolysis was accomplished without a cerebrospinal fluid fistula. Postoperatively, the patient improved. The histological examination confirmed OLF [Figure 4]. The MRI taken 2 weeks later confirmed adequate canal decompression [Figure 5]. Three months later, the patient was able to ambulate with a walker.

**DISCUSSION**

About 80% of the ligamentum flavum is composed of elastic fibers, and 20% with collagen.\(^{[1,14,27,35,36,44,48]}\) Ossification of the ligamentum flavum (OLF) involves heterotopic ossification of this ligament.

**Incidence**

There are 69 previously reported cases of cervical OLF [Table 1],\(^{[1,6,7,14,27,37,44,48]}\) which most typically occur in the East Asian population, followed by Caucasians [Table 2]. Patients with cervical OLF are between 27 and 84 years of age (average 62), and are mostly males [Tables 3 and 4].

**Clinical picture**

Patients with cervical OLF often present late in the clinical course with cord compression/spondylosis resulting in chronic myeloradiculopathy and an evolving quadriparesis.\(^{[1,14,27,44]}\) Cervical OLF and ossification of the posterior longitudinal ligament (OPLL) rarely appear together (e.g. 9/55 cases reported).

**Imaging**

Lateral cervical plain radiographs may demonstrate OLF located between the bases of two spinal processes.\(^{[23]}\) However, the location and severity of OLF are better demonstrated on MRI and CT studies. On T2-weighted sagittal MRI, OLF may be isointense, hypointense, or both,\(^{[27,35,36,44]}\) and there may be an accompanying intramedullary hyperintense signal [Table 5]. On CT scans examination, OLF lesions are usually seen as ossified masses extending from the facet joint to the base of the spinal processes, either unilaterally or bilaterally.

**Treatment**

A decompressive laminectomy with excision of OLF may be warranted in symptomatic patients. The OLF dissection from the dura should be accomplished under the operating microscope to lyse adhesions. Li et al. described decompressive en-block laminectomy for removal of OLF.\(^{[24]}\) Epstein, in 1999, proposed posterior stabilization for multilevel cervical OLF following extensive laminectomy.\(^{[9]}\) Dural tears should largely be avoided routinely using an operating microscope. If they

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**Figure 1:** Cervical MRI: (a) T1-weighted sagittal image showing a heterointense epidural mass at the posterior aspect of the spinal cord at the C4-C5 level, a smaller one is visualized below this level. (b) T2-weighted sagittal image demonstrating a hypointense mass with an isointensity at the center of the mass. (c) It also reveals a significant reduction in the cervical canal diameter.

**Figure 2:** Cervical computed tomography (CT) scan. (a) Sagittal reconstructed CT showing a calcified mass between the spinal processes of C4 and C5 with canal compromise present at the site of the ligamentum flavum. (b) Axial CT demonstrated an oval-shaped calcified mass.
Table 1: Review of all patients with cervical OLF

| N  | Author(s), Year       | Year | Sex | Age | Affected level(s)          | Race    |
|----|-----------------------|------|-----|-----|-----------------------------|---------|
| 1  | Koizumi[26]           | 1962 | M   | 55  | C6-C7                       | Japanese|
| 2  | Kirita et al.[18]     | 1973 | F   | 27  | C6-C7, C7-T1                | Japanese|
| 3  | Nagashima[21]         | 1975 | M   | 64  | C6-C7                       | Japanese|
| 4  | Kaneda[14]            | 1977 | M   | 46  | C2-C3, C3-C4                | Japanese|
| 5  | Kamakura et al.[15]   | 1978 | F   | 61  | C5-C6, C6-C7                | Japanese|
| 6  | Kubota et al.[23]     | 1981 | M   | 39  | C2-C3                       | Japanese|
| 7  | Kubota et al.[23]     | 1981 | F   | 47  | C2-C3                       | Japanese|
| 8  | Ota et al.[24]        | 1982 | F   | 75  | C5-C6                       | Japanese|
| 9  | Fujiwara et al.[11]   | 1982 | M   | 72  | C4-C5, C5-C6                | Japanese|
| 10 | Minami et al.[26]     | 1985 | M   | 40  | C7-T1                       | Japanese|
| 11 | Seichi et al.[16]     | 1988 | M   | 70  | C6-C7                       | Japanese|
| 12 | Tanaka et al.[47]     | 1988 | M   | 70  | C6-C7                       | Japanese|
| 13 | Hoshida et al.[13]    | 1989 | M   | 55  | C2-C3                       | Japanese|
| 14 | Sato et al.[30]       | 1989 | F   | 70  | C4-C5                       | Japanese|
| 15 | Sato et al.[30]       | 1989 | F   | 72  | C4-C5, C6-C7                | Japanese|
| 16 | Shimada et al.[41]    | 1990 | M   | 51  | C2-C3                       | Japanese|
| 17 | Kobayashi et al.[19]  | 1991 | M   | 61  | C3-C4                       | Japanese|
| 18 | Nishiwada[22]         | 1992 | F   | 52  | C5-C6                       | Japanese|
| 19 | Sugimura et al.[45]   | 1992 | F   | 62  | C4-C5                       | Japanese|
| 20 | Takayama et al.[46]   | 1993 | M   | 55  | C3-C4, C4-C5                | Japanese|
| 21 | Doi et al.[38]        | 2000 | F   | 72  | C3-C4, C6-C7                | Japanese|
| 22 | Kruse et al.[22]      | 2000 | …   | 72  | C3-C4                       | American|
| 23 | Mizuno and Nakgawa[20] | 2002 | M   | 64  | C6-C7                       | Japanese|
| 24 | Li et al.[24]         | 2002 | M   | 47  | C3-C4                       | Chinese |
| 25 | Li et al.[24]         | 2002 | M   | 58  | C4-C5, C5-C6                | Chinese |
| 26 | Mak et al.[25]        | 2002 | M   | 71  | C7-T1                       | Chinese |
| 27 | Chou et al.[50]       | 2004 | F   | 40  | C2-C3                       | Chinese |
| 28 | Nadkarni et al.[100] | 2005 | M   | 30  | C1-C2                       | Indian  |
| 29 | Yang et al.[49]       | 2005 | F   | 57  | C4-C5, C5-C6, C6-C7         | Chinese |
| 30 | Fukao et al.[12]      | 2006 | M   | 72  | C2-C3                       | Japanese|
| 31 | Chen et al.[31]       | 2007 | F   | 61  | C4-C5                       | Chinese |
| 32 | Kim et al.[17]        | 2008 | F   | 58  | C3-C4                       | Japanese|
| 33 | Kim et al.[17]        | 2008 | M   | 63  | C3-C4, C4-C5                | Japanese|
| 34 | Singhal et al.[42]    | 2009 | F   | 50  | C2-C3                       | Indian  |
| 35 | Singhal et al.[42]    | 2009 | M   | 65  | C3-C4                       | Indian  |
| 36 | Ohnishi et al.[33]    | 2010 | M   | 74  | C3-C4                       | Japanese|
| 37 | Fotakopoulos et al.[10] | 2010 | F   | 84  | C4-C5, C5-C6                | Caucasian|
| 38 | Fotakopoulos et al.[10] | 2010 | F   | 72  | C4-C5                       | Caucasian|
| 39 | Yang et al.[49]       | 2011 | M   | 37  | C5-C6                       | Chinese |
| 40 | Yang et al.[44]       | 2011 | F   | 63  | C4-C5, C5-C6, C6-C7         | Chinese |
| 41 | Yang et al.[44]       | 2011 | M   | 62  | C6-C7                       | Chinese |
| 42 | Yang et al.[44]       | 2011 | F   | 50  | C4-C5, C5-C6                | Chinese |
| 43 | Yang et al.[44]       | 2011 | M   | 63  | C2-C3, C3-C4, C4-C5         | Chinese |
| 44 | Yang et al.[44]       | 2011 | F   | 75  | C3-C4, C4-C5, C5-C6         | Chinese |
| 45 | Yang et al.[44]       | 2011 | F   | 58  | C6-C7, C7-T1                | Chinese |
| 46 | Yang et al.[44]       | 2011 | M   | 65  | C4-C5                       | Chinese |
| 47 | Yang et al.[44]       | 2011 | F   | 74  | C3-C4, C4-C5, C5-C6         | Chinese |
| 48 | Yang et al.[44]       | 2011 | F   | 58  | C3-C4, C4-C5                | Chinese |
| 49 | Yang et al.[44]       | 2011 | M   | 45  | C6-C7                       | Chinese |
| 50 | Yang et al.[44]       | 2011 | M   | 58  | C5-C6, C6-C7                | Chinese |

Contd...
occur, closure with 7-0 Gortex sutures and microdural staples is warranted.

Outcome
Typically following a cervical laminectomy with resection of OLF, patients should significantly recover from their preoperative myeloradiculopathy.

Summary
Cervical OLF rarely causes cervical myeloradiculopathy. Following both MR and CT studies to adequately document the location/extent of disease, laminectomy alone often suffices to decompress the cord.

Table 1: Contd...

| N  | Author(s), Year | Year | Sex | Age | Affected level(s) | Race            |
|----|-----------------|------|-----|-----|-------------------|-----------------|
| 51 | Yang et al.[48] | 2011 | M   | 55  | C4-C5, C5-C6      | Chinese         |
| 52 | Yang et al.[48] | 2011 | F   | 62  | C6-C7, C7-T1      | Chinese         |
| 53 | Yang et al.[48] | 2011 | F   | 70  | C3-C4, C4-C5, C5-C6 | Chinese        |
| 54 | Yang et al.[48] | 2011 | M   | 50  | C2-C3             | Chinese         |
| 55 | Yang et al.[48] | 2011 | M   | 65  | C3-C4             | Chinese         |
| 56 | Yang et al.[48] | 2011 | M   | 50  | C5-C6             | Chinese         |
| 57 | Yang et al.[48] | 2011 | M   | 42  | C2-C3, C3-C4, C4-C5 | Chinese        |
| 58 | Dewachter et al.[7] | 2011 | F   | 67  | C6-C7             | Caucasian       |
| 59 | Christiano et al.[8] | 2011 | F   | 45  | C2…T1            | Hispanic        |
| 60 | Mohindra et al.[29] | 2011 | M   | 35  | C7-D3             | Indian          |
| 61 | Song et al.[43] | 2012 | M   | 50  | C5-C6             | Korean          |
| 62 | Inoue et al.[14] | 2013 | M   | 42  | C2-C3, C3-C4, C4-C5 | Japanese         |
| 63 | Kotani et al.[21] | 2013 | M   | 76  | C3-C4             | Japanese         |
| 64 | Kotani et al.[21] | 2013 | M   | 75  | C2-C3             | Japanese         |
| 65 | Shepard et al.[40] | 2015 | F   | 35  | C7-T1             | African-American |
| 66 | Chachan et al.[2] | 2016 | M   | 69  | C5-C6             | Singaporean      |
| 67 | Sampanis et al.[37] | 2016 | M   | 45  | C2-C3, C4-C5      | Caucasian         |
| 68 | Chitoku et al.[4] | 2017 | M   | 69  | C7-T1             | Japanese         |
| 69 | Current case    | 2018 | M   | 81  | C3-C4, C4-C5      | Caucasian         |

Table 2: The ethnicity of the patients with cervical OLF

| Race          | Frequency | Percent |
|---------------|-----------|---------|
| East Asian    | 58        | 84.1    |
| Caucasian     | 5         | 7.2     |
| Indian        | 4         | 5.8     |
| African-American | 1   | 1.4     |
| Hispanic      | 1         | 1.4     |
| Total         | 69        | 100.0   |

Table 3: The gender of the patients with cervical OLF

| Gender      | Frequency | Percent |
|-------------|-----------|---------|
| Unreported  | 1         | 1.4     |
| Female      | 28        | 40.6    |
| Male        | 40        | 58.0    |
| Total       | 69        | 100.0   |

Table 4: (a) The range of the age and the mean of the patients with cervical OLF

| n  | Minimum | Maximum | Mean | Std. deviation |
|----|---------|---------|------|----------------|
| Age| 69      | 27      | 84   | 13.058         |

(b) Descriptive analysis for the distribution of patients in different age groups

| Age group         | Frequency | Percent |
|-------------------|-----------|---------|
| Young adults      | 4         | 5.8     |
| Middle-aged adults| 19        | 27.5    |
| Older adults      | 46        | 66.7    |
| Total             | 69        | 100.0   |

Table 5: The frequency of OLF in different cervical levels

| Level   | Frequency | Percent |
|---------|-----------|---------|
| C1-C2   | 1         | 0.9     |
| C2-C3   | 15        | 13.9    |
| C3-C4   | 21        | 19.4    |
| C4-C5   | 25        | 23.1    |
| C5-C6   | 20        | 18.5    |
| C6-C7   | 17        | 15.7    |
| C7-T1   | 9         | 8.3     |
| Total   | 108       | 100.0   |

Declaration of patient consent
Written informed consent was obtained from the patient for publication and corresponding images.
Figure 3: The surgical specimen shows the calcified mass that is almost removed en-block.

Figure 4: Histological examination of the surgical specimen (a) and (b) shows areas of endochondral ossification or new bone formation. At the edge, elastic bundles compatible with ligaments are noted.

Figure 5: Post-op sagittal cervical MRI, (a) and (b) laminectomy and decompression of the cord is shown both in T1- and T2-weighted images.

Contribution
The steps of this article from design to writing were made by Abolfazl Rahimizadeh, Naser Asgari, Housain Soufiani, and Shaghayegh Rahimizadeh, retrospectively.

Ethical approval
Approved.

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Conflicts of interest
There are no conflicts of interest.

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