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Review Article

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

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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%).[27,44] In 1962, Koizumi described cervical OLF at autopsy in a 55-year-old male who had developed progressive quadriparesis.[20] Since then only 68 more cases of cervical OLF have been published, typically involving the East Asian populations presenting varying degrees of myelopathy.[1,4,6,7,9,14,20,21,27,37,40,44,48] 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

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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.

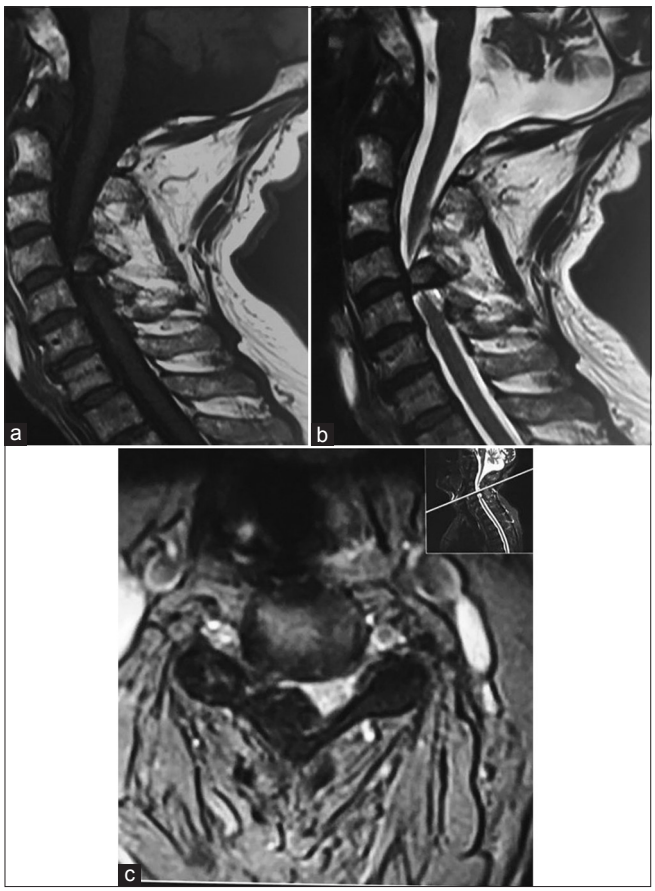

Figure 1: Cervical MRI: (a) T1-weighted sagittal image showing a heterointense epidural mass at the posterior aspect of the spinal cord at the C4-C5 level, a smaller one is visualized below this level. (b) T2-weighted sagittal image demonstrating a hypointense mass with an isointensity at the center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

Incidence

There are 69 previously reported cases of cervical OLF [Table 1],[1,4,6,7,14,27,37,44,48] 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

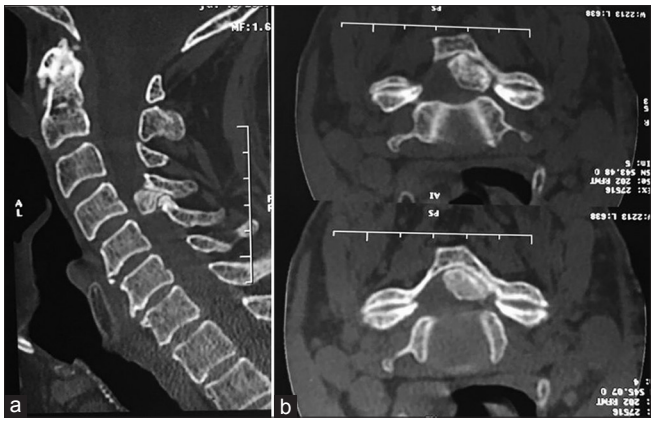

Figure 2: Cervical computed tomography (CT) scan. (a) Sagittal reconstructed CT showing a calcified mass between the spinal processes of C4 and C5 with canal compromise present at the site of the ligamentum flavum. (b) Axial CT demonstrated an oval-shaped calcified mass

Table 1: Review of all patients with cervical OLF

N	Author(s), Year	Year	Sex	Age	Affected level(s)	Race
1	Koizumi[20]	1962	M	55	C6-C7	Japanese
2	Kirita *et al*.[18]	1973	F	27	C6-C7, C7-T1	Japanese
3	Nagashima[31]	1975	M	64	C6-C7	Japanese
4	Kaneda[16]	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*.[34]	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*.[39]	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*.[38]	1989	F	70	C4-C5	Japanese
15	Sato *et al*.[38]	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	Nishiura[32]	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*.[8]	2000	F	72	C3-C4, C6-C7	Japanese
22	Kruse *et al*.[22]	2000	…	72	C3-C4	American
23	Mizuno and Nakgawa[28]	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*.[5]	2004	F	40	C2-C3	Chinese
28	Nadkarni *et al*.[30]	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*.[3]	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*.[48]	2011	M	37	C5-C6	Chinese
40	Yang *et al*.[48]	2011	F	63	C4-C5, C5-C6, C6-C7	Chinese
41	Yang *et al*.[48]	2011	M	62	C6-C7	Chinese
42	Yang *et al*.[48]	2011	F	50	C4-C5, C5-C6	Chinese
43	Yang *et al*.[48]	2011	M	63	C2C3, C3-C4, C4-C5	Chinese
44	Yang *et al*.[48]	2011	F	75	C3-C4, C4-C5, C5-C6	Chinese
45	Yang *et al*.[48]	2011	F	58	C6-C7, C7-T1	Chinese
46	Yang *et al*.[48]	2011	M	65	C4-C5	Chinese
47	Yang *et al*.[48]	2011	F	74	C3-C4, C4-C5, C5-C6	Chinese
48	Yang *et al*.[48]	2011	F	58	C3-C4, C4-C5	Chinese
49	Yang *et al*.[48]	2011	M	45	C6-C7	Chinese
50	Yang *et al*.[48]	2011	M	58	C5-C6, C6-C7	Chinese

Contd...

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	F	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*.[6]	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

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

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

Outcome

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

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 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	58.62	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

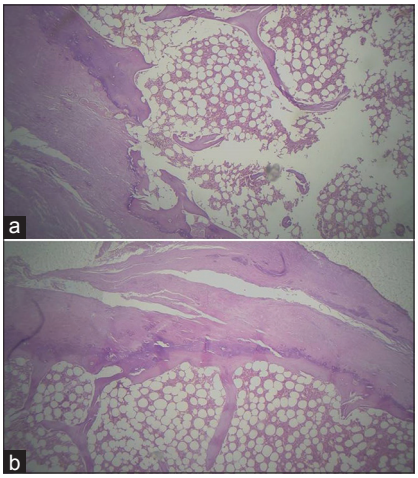

Figure 4: Histological examination of the surgical specimen (a) and (b) shows areas of endochondral ossification or new bone formation. At the edge, elastic bundles compatible with ligaments are noted

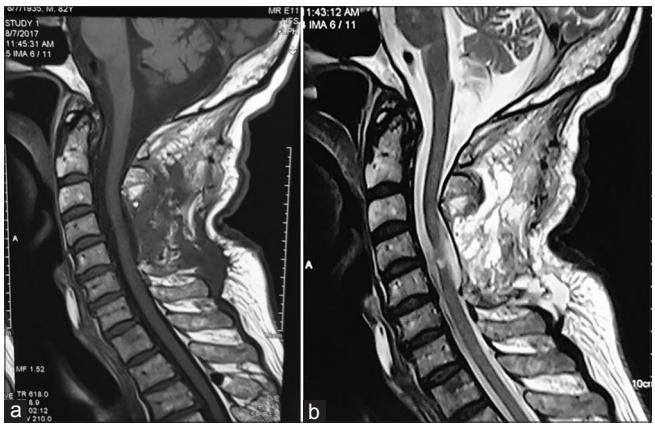

Figure 5: Post-op sagittal cervical MRI, (a) and (b) laminectomy and decompression of the cord is shown both in T1- and T2-weighted images

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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Nil.

Conflicts of interest

There are no conflicts of interest.

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