

Radiological manifestations and surgical outcome of combined upper cervical cord compression and cervical ossification of the posterior longitudinal ligament with a minimum 2-year follow-up

Hao Li, MD^a, Xiaopeng Zhou, MD^a, Gang Chen, MD^a, Fangcai Li, MD, PhD^a, Junfeng Zhu, MD^b, Qixin Chen, MD, PhD^{a,*}

Abstract

Combined upper cervical cord compression associated with cervical ossification of the posterior longitudinal ligament (OPLL) is a rare and under-recognized disorder. The aim of this study was to investigate the radiological manifestations and surgical outcome of this combined disease.

Between May 2011 and July 2015, patients who underwent surgery for combined upper cervical cord compression and cervical OPLL in our institution were included in this study. After a minimum 2-year follow-up, radiological and clinical data were collected. The etiology of upper cervical cord compression and radiological features of cervical OPLL was determined. Surgical outcome was evaluated with Visual Analogue Scale (VAS), Japanese Orthopedic Association score (JOA), space available for the spinal cord (SAC) at the cephalad adjacent level, occupying ratio of OPLL and cervical lordosis.

In total, 24 patients (11 men and 13 women) with a mean age of 57.9 years old were included. The etiology of upper cervical cord compression included craniovertebral junction deformity (n = 10), atlantoaxial subluxation (n = 5), and OPLL extending to C2 level (n = 9). The extent, type, and thickest level of cervical OPLL varied among the patients. Significant improvement of VAS and JOA score was noted postoperatively and at a minimum 2-year follow-up. The result was satisfactory in SAC at the cephalad level and occupying ratio of OPLL. There were no significant differences in C2/C7 lordotic angle at the preoperative, postoperative and the last follow-up examination.

In conclusion, the radiological manifestations of combined upper cervical cord compression and cervical OPLL varied among the patients. Satisfied results can be achieved with adequate surgical treatment a minimum 2-year follow-up.

Abbreviations: CT = computed tomography, JOA = Japanese Orthopedic Association score, MRI = magnetic resonance imaging, OPLL = cervical ossification of the posterior longitudinal ligament, SAC = space available for the spinal cord, VAS = Visual Analogue Scale.

Keywords: ossification of the posterior longitudinal ligament, radiological manifestation, surgery, upper cervical cord compression

Editor: Zelena Dora.

This work was supported by the grants from the National Nature Science Foundation of China (No. 81501908) and Science and Technology Planning Project of Zhejiang Province (No. 2017KY076).

The authors have no conflicts of interest to disclose.

^a Department of Orthopedics, 2nd Affiliated Hospital, School of Medicine, Zhejiang University, Hangzhou, ^b Department of Orthopedics, Suichang People's Hospital, Lishui, P.R. China.

* Correspondence: Qixin Chen, Department of Orthopedics, 2nd Affiliated Hospital, School of Medicine, Zhejiang University, 88 Jie fang Road, Hangzhou 310009, P.R. China (e-mail: zrcqx@zju.edu.cn).

Copyright © 2017 the Author(s). Published by Wolters Kluwer Health, Inc. This is an open access article distributed under the terms of the Creative Commons Attribution-Non Commercial License 4.0 (CCBY-NC), where it is permissible to download, share, remix, transform, and buildup the work provided it is properly cited. The work cannot be used commercially without permission from the journal.

Medicine (2017) 96:45(e8332)

Received: 19 April 2017 / Received in final form: 6 September 2017 / Accepted: 8 September 2017

<http://dx.doi.org/10.1097/MD.0000000000008332>

1. Introduction

Cervical cord compression is commonly present in elderly adulthood, often with associated degenerative disc disease, ossification of the posterior longitudinal ligament (OPLL), or diffuse idiopathic skeletal hyperostosis.^[1–3] Due to the relatively wide canal diameter, we rarely encounter spinal cord compression at the upper cervical segment (C1 and C2 levels, atlantoaxial and atlanto-occipital joints).^[4,5] Until now, only a few cases of upper cervical cord compression have been reported.^[6,7]

OPLL is a condition of ectopic bone formation within the posterior longitudinal ligament, which may cause cervical canal stenosis and cervical myelopathy.^[8,9] In recent years, an increasing number of reports have been published concerning OPLL with the growing recognition of the disease.^[10] In most cases, cervical OPLL is observed at C4 and C5 levels.^[11,12] OPLL at the C1 and C2 segments is a rare occurrence, and only a handful of cases have been documented in the literature.^[7,13,14]

It is extremely rare that upper cervical cord compression and cervical OPLL occur simultaneously in the same patient.

Therefore, the radiographic manifestations, as well as the surgical outcome, remain unclear for this uncommon and combined disease. In this study, we reported a case series of combined upper cervical cord compression and cervical OPLL triggering myelopathy. The objective of this study was to clarify the radiographic manifestations of combined upper cervical cord compression and cervical OPLL; evaluate the surgical outcome by determining the patients' symptoms relief and the changes of the diameter of cervical canal and cervical lordosis.

2. Methods

Between May 2011 and July 2015, 24 patients who underwent surgery for combined upper cervical cord compression and cervical OPLL were included in this study. Inclusion criteria of this study were more than 18 years old; X-ray and computed tomography (CT) of the cervical spine demonstrated cervical OPLL, CT and magnetic resonance imaging (MRI) demonstrated the sagittal diameter of cervical canal was ≤ 14 mm at the upper cervical segment (C1 and C2) or atlantoaxial, atlanto-odontoid and atlanto-occipital joints instability; surgery was performed for cervical myelopathy which was resistant to conservative therapy; a minimum of 2-year follow-up. Exclusion criteria were patients with history of previous trauma or surgery of the craniocervical and cervical regions; patients with incomplete X-ray, CT, and MRI data.

This study was approved by the Ethics Committee of the Second Affiliated Hospital, School of Medicine, Zhejiang University (Hangzhou, China). All the patients provided free written informed consent. Research was conducted in accordance with the research principles in the Declaration of Helsinki.

2.1. Radiological measurements

Radiographic examinations were first performed by a registered radiographer with 15 years of experience. A second radiographer with 21 years of experience repeated the measure to ensure the correctness and credibility of the result. Imaging studies included plain films, CT and MRI of the cervical spine. Based on CT and MRI examination, the etiology of upper cervical cord compression was determined. On the lateral plain films and sagittal CT scans, the type of OPLL was classified as continuous, segmental, mixed, and circumscribed type according to the report of the Investigation Committee on OPLL of the Japanese Ministry of Public Health and Welfare (now the Japanese Ministry of Health, Labour, and Welfare).^[15] Continuous type is classified as a long lesion extending over several vertebral bodies; segmental type is classified as 1 or several separate lesions behind vertebral bodies; mixed type is classified as a combination of continuous and segmental types; circumscribed type is classified as the lesion mainly located posterior to a disc space. On CT scan, the extent and the thickest level of OPLL were also identified.

2.2. Surgical methods

The included patients were stratified into 3 groups based on the etiologies of upper cervical cord compression and the corresponding procedures were performed by a highly skilled board-certified spine surgeon with 30 years of experience in treatment of spinal disorders. Group 1, craniocervical junction deformity and cervical OPLL (n=10): transoral decompression, posterior occipitoaxial fusion with open-door laminoplasty was performed in 6 cases of occipitalization of atlas and basilar invagination; resection of the posterior arch of C1 combined with open-door

laminoplasty was performed in 4 cases of hypoplasia of posterior arch of atlas. Group 2, atlantoaxial subluxation and cervical OPLL (n=5): atlantoaxial fixation, fusion combined with open-door laminoplasty, was performed as the atlantoaxial joint not to be reduced in extension position. Group 3, OPLL extending to C2 level (n=9): C2 dome-like laminoplasty combined with lower-level open-door laminoplasty at downward levels was performed.

2.3. Surgical outcome evaluation

We reviewed the medical records of the patients and obtained data regarding the demographic characteristics, neurological symptoms, and signs of the patients. For clinical evaluation, Japanese Orthopedic Association (JOA) scores for cervical myelopathy and Visual Analogue Scale (VAS) for neck pain were documented preoperatively, 3 months postoperatively and at the last follow-up. The recovery rate of JOA score was also used to assess the surgical outcome and was calculated as follows: recovery rate (%) = (the last follow-up JOA score – preoperative JOA score) / (17 – preoperative JOA score) $\times 100\%$. The recovery rate was ranked as excellent (75–100%), good (50–74%), fair (25–49%), poor (0–24%), or worse (<0) at the last follow-up. And all surgery-related complications were recorded postoperatively and at the follow-up examinations.

On coronal CT scan, the space available for the spinal cord (SAC) at the cephalad adjacent level was measured preoperatively, 3 months postoperatively and at the last follow-up. The occupying ratio of OPLL was measured as the diameter of the greatest thickness of OPLL divided by the anteroposterior diameter of the bony spinal canal at the same level on the CT axial imaging. Based on the lateral radiographs at the neutral position, global cervical lordosis (C2–C7 Cobb angle) was calculated by Cobb method.

2.4. Statistical analysis

A statistical analysis was performed using the paired *t* test for parameter preoperatively, postoperatively and at last follow-up. Significance was set at the level of $P < .05$ and SPSS software version 17.0 (IBM, Armonk, New York) was used. Results are presented as the mean \pm standard deviation.

3. Results

Finally, 24 patients (11 men and 13 women) at a mean age of 57.9 ± 12.8 years old (range, 36–88 years) were included in the study. Comorbidity included hypertension in 4 patients, rheumatoid arthritis in 2 patients, type 2 diabetes mellitus in 2 patients, and coronal heart disease in 1 patient.

3.1. Radiological manifestations

In the included 24 patients, upper cervical cord compression aroused from craniocervical junction deformity in 10 cases (41.7%), atlantoaxial subluxation in 5 cases (20.8%), and OPLL extending to C2 level in 9 cases (37.5%) (Table 1). The extent of cervical OPLL varied among the included patients, and the mixed type of OPLL was observed in 14 cases (58.3%), continuous type in 7 cases (29.2%), segmental type in 2 cases (8.3%), and circumscribed type in 1 case (4.2%). OPLL with greatest thickness was at the C5 level in 9 cases (37.5%), followed by C4 in 8 cases (33.3%), C3 in 6 cases (25.0%), and C4/C5 in 1 case (4.2%).

Table 1
The characteristics and radiological manifestations of the patients.

NO.	Age, y	Gender	FU, mo	Upper cervical cord compression	Group	OPLL		
						Extent	Type	Thickest level
1	63	M	27	Occipitalization of atlas, basilar invagination	1	C4/C5, C5/C6	Circumscribed	C4/C5
2	77	M	49	Occipitalization of atlas, basilar invagination	1	C3–C5, C6	Mixed	C5
3	56	F	27	OS odontoideum	1	C2–C6	Continuous	C4
4	55	F	36	Anomaly of odontoid process	1	C3–5, C6	Mixed	C5
5	55	F	24	Malunion of odontoid process	1	C3–4, C6	Mixed	C3
6	47	F	48	Occipitalization of the atlas, fusion of atlantoaxial facets	1	C2–C4, C6	Mixed	C3
7	71	F	26	Atlas hypoplasia	1	C3–C7	Mixed	C5
8	72	M	31	Atlas hypoplasia	1	C2–C7	Continuous	C3
9	63	F	43	Atlas hypoplasia	1	C4, C6	Segmental	C4
10	50	F	30	Atlas hypoplasia	1	C3–C4, C6	Mixed	C3
11	56	F	26	Atlantoaxial subluxation	2	C3–6	Mixed	C5
12	45	M	42	Atlantoaxial subluxation	2	C3–6	Continuous	C5
13	49	F	24	Atlantoaxial subluxation	2	C3–5, C6	Mixed	C4
14	68	F	25	Atlantoaxial subluxation	2	C3, C4, C7	Segmental	C4
15	45	M	31	Atlantoaxial subluxation	2	C3–C5, C6	Mixed	C5
16	41	F	34	OPLL extending to C2 level	3	C2–C5	Continuous	C4
17	36	F	48	OPLL extending to C2 level	3	C2–C6	Continuous	C5
18	44	M	30	OPLL extending to C2 level	3	C2–C5	Continuous	C5
19	68	M	36	OPLL extending to C2 level	3	C2–C4, C6	Mixed	C3
20	88	M	24	OPLL extending to C2 level	3	C2–C6	Continuous	C4
21	72	F	28	OPLL extending to C2 level	3	C2–C3, C5	Mixed	C4
22	49	M	24	OPLL extending to C2 level	3	C2–C6	Mixed	C5
23	60	M	25	OPLL extending to C2 level	3	C2–C4, C6	Mixed	C3
24	59	M	34	OPLL extending to C2 level	3	C2–C4, C5	Mixed	C4

F=female, FU=follow-up, M=male, OPLL=ossification of the posterior longitudinal ligament.

3.2. Surgical outcome

The mean follow-up time was 32.2 ± 8.2 months (range: 24–49 months). The mean VAS score was 5.9 ± 1.2 preoperatively, 2.6 ± 1.1 (P < .001) at 3-month postoperatively and 2.1 ±

0.9 (P < .001) at the last follow-up, respectively (Table 2). The mean JOA score was 10.3 ± 2.2 preoperatively, and it increased to 14.0 ± 2.1 (P < .001) at 3-month postoperatively and 14.4 ± 2.3 (P < .001) at the last follow-up. According to

Table 2
The surgical outcome of all patients.

NO.	VAS			JOA			OR of OPLL, %			SAC, mm			Cervical lordosis,°			Complication
	Pre	Post	Final	Pre	Post	Final	Pre	Post	Final	Pre	Post	Final	Pre	Post	Final	
1	8	5	3	9	10	10	63.3	27.0	29.9	7.1	15.8	15.4	18.1	18.4	19.6	Superficial infection
2	7	4	2	11	14	16	50.2	46.1	49.0	11.1	19.8	18.9	10.9	11.5	10.2	
3	7	3	1	7	12	14	51.3	25.1	28.6	10.3	18.6	18.6	15.3	16.6	17.4	
4	6	2	3	8	14	15	55.8	37.2	44.2	9.9	19.3	17.9	11.2	11.0	10.6	
5	5	4	3	9	9	9	71.4	38.2	42.0	11.3	20.1	20.2	17.1	18.8	19.2	
6	8	4	4	7	15	15	57.2	28.8	33.3	11.1	18.3	15.9	14.9	15.9	16.8	
7	6	3	2	10	12	10	56.7	25.2	28.3	10.3	20.2	20.0	14.6	18.3	18.6	
8	6	3	2	7	14	15	64.8	34.3	33.4	12.9	21.3	21.0	6.7	8.9	9.2	
9	7	2	3	9	12	15	61.1	30.5	30.6	11.6	18.9	18.9	12.7	15.3	15.4	
10	8	4	2	10	15	16	49.4	26.3	30.1	12.3	20.4	20.0	17.6	16.8	19.0	
11	5	3	2	10	16	16	31.3	19.2	23.6	12.3	20.9	18.8	18.7	20.0	20.5	
12	6	2	1	10	15	15	49.3	19.6	31.2	13.3	19.0	19.6	19.5	17.3	16.8	
13	7	3	2	14	16	17	43.6	28.9	26.2	11.2	20.8	19.5	12.8	10.3	13.9	
14	6	2	2	12	16	16	66.4	20.3	29.9	13.6	20.1	21.0	20.4	19.4	19.6	
15	5	1	1	11	14	14	51.6	27.1	25.5	9.9	19.9	17.7	16.2	16.0	19.5	
16	4	1	1	10	12	11	48.5	20.6	29.1	17.6	18.1	18.6	–5.8	–4.0	–1.5	
17	5	3	2	11	14	15	42.3	17.7	16.9	16.5	18.3	17.2	–8.3	–8.9	–10.4	
18	4	2	1	12	12	12	61.2	25.6	31.6	16.3	17.9	18.8	4.9	6.3	6.9	
19	5	2	3	14	15	15	76.5	27.5	21.7	17.3	18.5	18.5	10.9	8.6	7.9	C5 palsy
20	6	2	2	15	16	16	47.3	22.9	33.3	16.9	19.1	18.8	–7.6	–7.0	–5.5	
21	5	1	2	12	14	15	56.2	23.6	23.9	14.3	20.4	20.3	2.2	–1.0	–5.8	C5 palsy
22	4	2	1	7	17	17	65.7	19.0	19.6	18.1	21.3	19.9	–6.1	–5.5	–5.6	
23	7	4	4	10	16	16	70.2	21.7	16.8	16.4	16.9	17.1	15.4	15.4	10.7	
24	5	1	1	11	16	15	42.2	19.8	20.8	17.2	19.8	17.9	14.6	13.0	12.3	

JOA=Japanese Orthopedic Association, OR=occupying ratio, Post=3-Mo postoperative, Pre=preoperative, SAC=space available for the spinal cord, VAS=Visual Analogue Scale.



Figure 1. Patient 1 (shown in Table 1) was a 63-year-old man with occipital-atlanto abnormalities and cervical OPLL. This patient underwent transoral decompression, posterior C1 laminectomy, and occipitoaxial fusion using iliac crest bone graft with C3–C6 open-door laminoplasty. A, Preoperative lateral X-ray showed fusion of the atlanto–occipital joints. B, Preoperative sagittal CT demonstrated occipitalization of the atlas, the tip of the odontoid projects more than 3mm above Chamberlain line, basilar invagination, circumscribed-type OPLL at C4/5 and C5/6. C, T2-weighted MRI demonstrated spinal cord compression at the level of occipital–atlanto junction and C2 to C6, with a high signal change at the level of occipital–atlanto junction. D, E, Postoperative sagittal and coronal CT showed the left C1 lamina was excised and satisfactory neural decompression and internal fixation was achieved. F, Lateral X-ray at 24 months postoperatively showing the satisfactory overall sagittal alignment of the cervical spine. CT = computed tomography, MRI = magnetic resonance imaging, OPLL = ossification of the posterior longitudinal ligament.

the recovery rate of JOA, 11 patients (45.8%) achieved excellent recovery, and 7 patients (29.2%) achieved good recovery at the last follow-up.

The preoperative mean occupying ratio of OPLL was $55.6\% \pm 10.8\%$ and the mean SAC at the cephalad adjacent level was 13.3 ± 3.1 mm. Postoperatively, the mean occupying ratio of OPLL decreased to $26.3\% \pm 7.0\%$ ($P < .001$) and SAC at the cephalad adjacent level increased to 19.3 ± 1.4 mm ($P < .001$). At the last follow-up, the mean occupying ratio of OPLL was $29.1\% \pm 7.9\%$ ($P < .001$) and SAC at the cephalad adjacent level was 18.8 ± 1.4 mm ($P < .001$). The mean preoperative C2–C7 Cobb angle was $10.3 \pm 9.0^\circ$, and it did not change significantly at 3 months postoperatively and at the last follow-up, as the Cobb angle was $10.5 \pm 9.1^\circ$ ($P = .60$) and $10.6 \pm 9.6^\circ$ ($P = .57$), respectively (Table 2 and Figs. 1–3).

During the follow-up, unilateral C5 palsy was observed in 2 patients and completely recovered 7 and 11 months later after the surgery, respectively. And 1 patient with diabetes mellitus type 2 experienced superficial infection, which was successfully treated by antibiotic administration and twice wound debridement. No cerebrospinal fluid leak or pulmonary embolism was observed in any patient. No complication of internal fixation was observed during the follow-up, and bone union was achieved following bone grafting in all fusion cases.

4. Discussion

As symptomatic upper cervical cord compression and cervical OPLL occur simultaneously is quite uncommon, there are only 12 studies (50 cases) reported in the English literature till today^[6,7,13,16–24] (Table 3). Thus, the radiographic manifestations, as well as the surgical outcome of this disease, remain unclear. In this study, a case series of cervical myelopathy due to the concurrence of upper cervical cord compression and cervical OPLL was studied. A detailed investigation of this case series revealed that the radiological manifestations of combined upper cervical cord compression and cervical OPLL varied among the patients; satisfied results can be achieved with adequate surgical treatment at a minimum 2-year follow-up.

The radiological manifestations and pathology of combined upper cervical cord compression and cervical OPLL were discussed in the previous studies, though it has not been well understood and defined yet. Some authors thought OPLL is a potential factor contributing to the development of atlantoaxial lesions. For example, Kawabori et al^[21] reported a rare case of DISH with continuous type of OPLL at C2–4 levels and C-1 posterior tubercle impingement on the spinal cord that presented with cervical myelopathy. They proposed that fusion of the subaxial cervical spine caused the hypermobility at C1–2, and may lead to the ligamentous damage and upper cervical cord

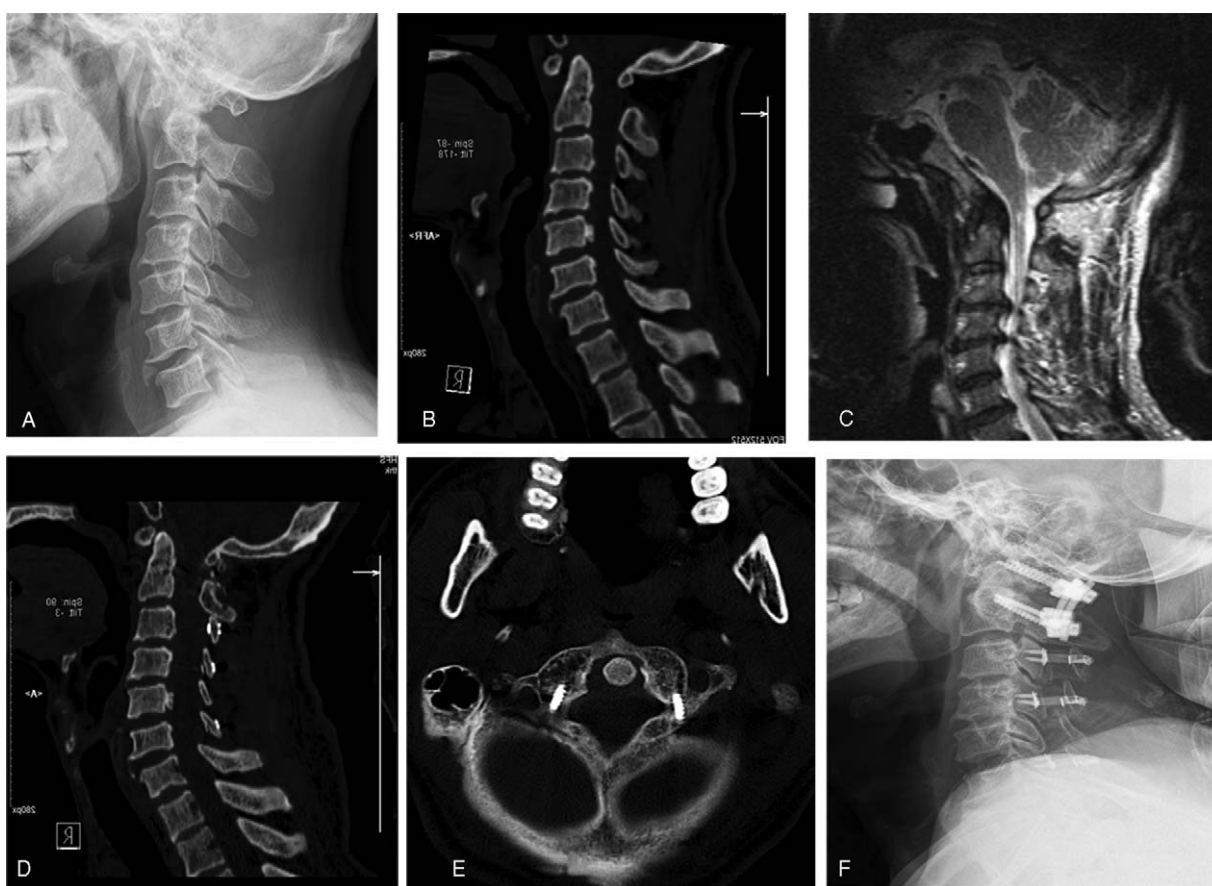


Figure 2. Patient 11 (shown in Table 1) was a 56-year-old man with atlantoaxial subluxation and cervical OPLL. This patient underwent atlantoaxial fixation, fusion with C3–C6 open-door laminoplasty. A, Preoperative lateral X-ray showed atlantoaxial subluxation and segmental-type OPLL from C3 to C6. B, Preoperative sagittal CT demonstrated the cervical spinal stenosis at the level of atlas and axis and segmental-type OPLL from C3 to C6. C, T2-weighted MRI demonstrated spinal cord compression at C1–C6 with a high signal change at C1 level. D, E, Postoperative sagittal and coronal CT showed subluxation was reduced and satisfactory neural decompression and internal fixation. F, Lateral X-ray at 26 months postoperatively showing the satisfactory overall sagittal alignment of the cervical spine. CT = computed tomography, MRI = magnetic resonance imaging, OPLL = ossification of the posterior longitudinal ligament.

compression. In another study, Takasita et al^[19] suggested that OPLL can affect the cervical biomechanics and restricts the mobility of the lower cervical spine, which causes additional mechanical demand on the atlantoaxial joint and subsequently leads to atlantoaxial subluxation. However, some authors disagreed with this hypothesis and proposed the pathology of combined upper cervical cord compression and cervical OPLL is that upper cervical cord compression occasionally occurred in patients with cervical OPLL. For example, Shirado et al^[20] reported a case of cervical myelopathy due to combined atlantoaxial subluxation and cervical OPLL. They thought it is just a rare coincidence that cervical OPLL occasionally occurred in a rheumatoid arthritis patient with atlantoaxial subluxation, which resulted in cervical myelopathy. In this study, we found that the etiology of combined upper cervical cord compression and the radiological features of cervical OPLL varied among the patients. This finding supports the idea that it is just a rare coincidence that upper cervical cord compression and cervical OPLL occasionally occurred in the same patient.

The goal of surgical treatment for combined upper cervical cord compression and cervical OPLL is eliminating spinal cord compression and stopping the neurologic deterioration.^[25,26] Currently, no definite surgical strategy has been established. As

we know, the treatment strategy for upper cervical spinal lesions should consider the occurrence of spinal cord compression or spinal instability, or both. As to the treatment of cervical OPLL, the mainstay treatment is surgical decompression.^[9] Both the anterior and posterior operative procedures for upper cervical cord compression and cervical OPLL have been shown effective in both clinical and imaging outcomes. It is reported that anterior approach is associated with better overall postoperative neural function and effective for the decompression of massive OPLL and with cervical kyphosis.^[27,28] However, the complex anatomic structures around the upper cervical spine involving the C2 segment make it difficult to perform anterior surgery given the high complication rates, even for skilled surgeons.^[29] According to the study of Liu et al, the surgery-related complication rate for multilevel cervical myelopathy was nearly 37.3% using the anterior approach. On the other hand, posterior surgical approach is safer and more effective regarding the development of surgery-related complications.^[30,31] But the posterior surgical approach is associated with the risk of postoperative kyphosis as a result of paraspinal muscle injury at C2 segment, postoperative axial neck pain, and symptomatic aggravation due to the progression of OPLL. Consequently, it is not easy to choose an effective surgical approach and determine

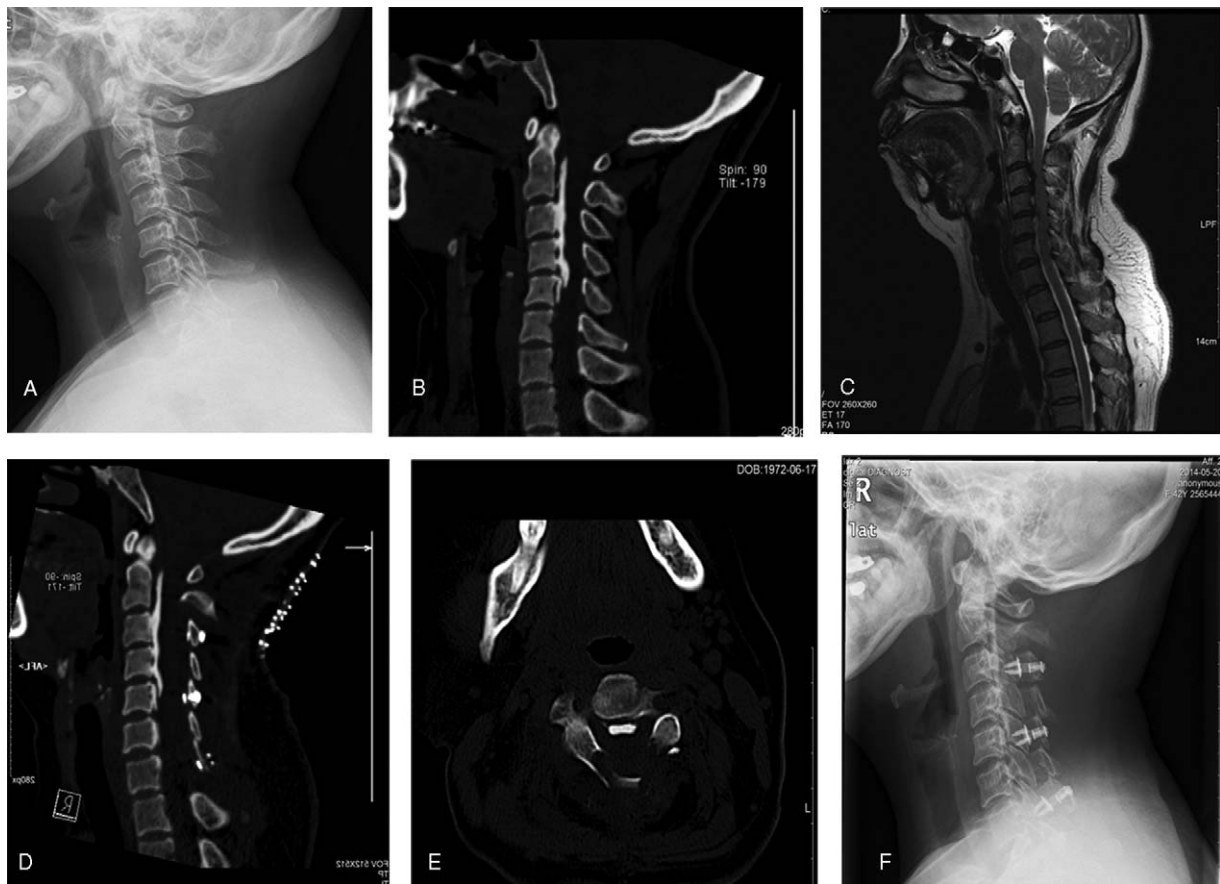


Figure 3. Patient 16 (shown in Table 1) was a 41-year-old woman with cervical OPLL extending to C2 level that underwent C2 dome-like laminoplasty with C3–C7 open-door laminoplasty. A, Preoperative lateral X-ray showed the OPLL from C2 to C6. B, Preoperative sagittal CT demonstrated OPLL extending to C2 level, resulting cervical stenosis. C, T2-weighted MRI demonstrated spinal cord compression at C2–C7. D, E, Postoperative sagittal and coronal CT showed a dome-like excision of the inner side of C2 spinal canal and satisfactory neural decompression and internal fixation from C2 to C7. F, Lateral X-ray at 34 months postoperatively showing the satisfactory overall sagittal alignment of the cervical spine.

the surgical extent in cases of cervical OPLL combined with upper cervical cord compression.

In summary, the surgery for combined upper cervical cord compression and cervical OPLL must be weighed against the type and severity of stenosis, the type and extent of OPLL, sagittal alignment, and history of previous surgery.

Similarly to the previous related studies, all surgeries were performed using the posterior approach in the present study. The reasons include: it is difficult to perform cranial decompression at C2 segment through anterior approach; the coexist OPLL at C3 and downward segments also need decompression; decompression should be performed at 3 or more levels in most cases, but anterior approach is associated with high rates of pseudarthrosis and dysphagia when 3 or more levels require treatment; an anterior approach was deemed risky, as it requires technique-related skills and the chance of the development of complications is high. And in our opinion, posterior fusion should be mandatory for patients with postdecompression spinal instability, because single decompression may aggravate the instability of the upper cervical spine, thereby leading to neurological deterioration. In general, we categorized the patients into 3 groups based on the pathology of upper cervical cord compression. Finally, we

performed adequate posterior decompression for patients in the 3 groups, respectively.

As to the surgical outcomes of combined upper cervical cord compression and cervical OPLL, a minimum 2-year follow-up result indicated that the neurological function improved in most patients as the JOA scores significantly increased and 11 patients (45.8%) achieved excellent recovery, and 7 patients (29.2%) achieved good recovery at the last follow-up. In addition, no patients developed progressive kyphosis or progression of ossified lesions during the follow-up. A similar result was noted in all the previous studies, as most cases achieved excellent clinical outcomes with low complication after posterior surgeries. These results confirmed the significance of surgical treatment for combined upper cervical cord compression and cervical OPLL.

There are several limitations to the present study that warrant mention: small sample size, retrospective study, not including conservative managed patients, all which might have caused substantial bias. More multicenter studies and prospective studies are needed to create studies with high levels of evidence.

In conclusion, the results of this study showed that in patients with combined upper cervical cord compression and cervical OPLL, the etiology of upper cervical cord compression included

Table 3
Reported cases of combined upper cervical cord compression and cervical OPLL.

Author and year	No. of cases	Age (y)/gender	Upper cervical cord compression	OPLL extent	OPLL type	Surgical procedure	FU (mo)	Pre/final neurologic score/tool	Clinical result
Kubota et al (1981) ^[16]	1	47 F	C2-C3 OLF	C2-C4	Continuous	C2-C6 laminectomy, C2-C6 dome-like laminoplasty and downward laminoplasty	8	NM	Improved
Matsuzaki et al (1989) ^[17]	25	NM	OPLL extending to C2 level	C2-T1	NM	Atlas laminectomy, axis dome-like laminectomy, C3-C6 laminectomy and occipitoxial fusion	Range: 12-84	NM	Improved in all case
Chiba et al (1992) ^[18]	1	55 M	Atlantoaxial Subluxation	C2-C7	Continuous	Atlas laminectomy, axis dome-like laminectomy, C3-C6 laminectomy and occipitoxial fusion	28	7/14/JOA	Improved
Takasita et al (2000) ^[19]	2	74 F	Atlantoaxial subluxation	C2-C6	Mixed	Atlas laminectomy, C3-C7 laminoplasty and occipitoxial arthrodesis	NM	NM	Improved
Shirado et al (2005) ^[20]	1	64 M 68 F	Atlantoaxial subluxation	C3-C5 C5-C7	Continuous Continuous	Atlas laminectomy, C3-C7 laminoplasty and occipitoxial fusion	60	NM	Improved Improved
Kawabori et al (2009) ^[21]	1	75 M	Atlas impingement	C2-C4	Continuous	C1-C3 laminectomy, C1 posterior arch resection	NM	NM	Improved
Takeuchi et al (2013) ^[6]	1	40 M	Atlas impingement	C2-C5	Mixed	C2-C3 laminectomy, C4-C6 laminoplasty	13	NM	Improved
Kotani et al (2013) ^[22]	1	75 M	C2-C3 OLF	C3-C7	Mixed	C2 laminectomy and C2-3 instrumented fusion	24	5/12/JOA	Improved
Passias et al (2013) ^[7]	1	49 M	Posterior axis abnormalities	C2-C3	Segmental	C2: 10 laminectomy and 4 laminoplasty; Below C2: 5 laminectomy and 9 laminoplasty	NM	11/14 /JOA	Improved
Lee et al (2015) ^[13]	14	13 M 1 F	OPLL extending to C2 level	C2-C7	3 Continuous Mixed	C2: 10 laminectomy and 4 laminoplasty; Below C2: 5 laminectomy and 9 laminoplasty	71.79±65.15	12.64/15.71 /mean JOA	67.58% Recovery rate
Shiba(2015) ^[23]	1	32 F	OPLL extending to C2 level	C2-C7	Mixed	C2 dome-like laminotomy, C3-C6 laminoplasty	14	7/14/JOA	57.1% Recovery rate
Hirao et al (2016) ^[24]	1'	65 F	OPLL extending to C1 level and occipital bone	Extensive	Continuous	C1 posterior arch and ossified rim of occipital bone resection	18	NM	Improved

F = female, FU = follow-up, JOA = Japanese Orthopedic Association, M = male, NM = not mention, OLF = ossification of ligamentum flavum, OPLL = ossification of the posterior longitudinal ligament, Pre = Preoperative.

craniovertebral junction deformity, atlantoaxial subluxation, and OPLL extending to C2 level; the radiological features of cervical OPLL varied. Satisfied results can be achieved with adequate surgical treatment at a minimum 2-year follow-up.

References

- [1] Mayr MT, Subach BR, Comey CH, et al. Cervical spinal stenosis: outcome after anterior corpectomy, allograft reconstruction, and instrumentation. *J Neurosurg* 2002;96(1 suppl):10–6.
- [2] Elsenbruch S. How positive and negative expectations shape the experience of visceral pain. *Handb Exp Pharmacol* 2014;225:97–119.
- [3] Liu FY, Ma L, Huo LS, et al. Mini-plate fixation versus suture suspensory fixation in cervical laminoplasty: a meta-analysis. *Medicine (Baltimore)* 2017;96:e6026.
- [4] Chanplakorn P, Kraiwattanapong C, Aroonjarattham K, et al. Morphometric evaluation of subaxial cervical spine using multi-detector computerized tomography (MD-CT) scan: the consideration for cervical pedicle screws fixation. *BMC Musculoskelet Disord* 2014;15:125.
- [5] Kasliwal MK, Traynelis VC. Hypertrophic posterior arch of atlas causing cervical myelopathy. *Asian Spine J* 2012;6:284–6.
- [6] Takeuchi M, Wakao N, Kamiya M, et al. Upper cervical cord compression due to a C-1 posterior arch in a patient with ossification of the posterior longitudinal ligament and a kyphotic cervical spine in the protruded-head position: case report. *J Neurosurg Spine* 2013;19:431–5.
- [7] Passias PG, Wang S, Wang S. Combined ossification of the posterior longitudinal ligament at C2-3 and invagination of the posterior axis resulting in myelopathy. *Eur Spine J* 2013;22(suppl 3):S478–86.
- [8] Liu Y, Zhao Y, Chen Y, et al. RUNX2 polymorphisms associated with OPLL and OLF in the Han population. *Clin Orthop Relat Res* 2010;468:3333–41.
- [9] An HS, Al-Shihabi L, Kurd M. Surgical treatment for ossification of the posterior longitudinal ligament in the cervical spine. *J Am Acad Orthop Surg* 2014;22:420–9.
- [10] Goel A, Nadkarni T, Shah A, et al. Is only stabilization the ideal treatment for ossified posterior longitudinal ligament? Report of early results with a preliminary experience in 14 patients. *World Neurosurg* 2015;84:813–9.
- [11] Sohn S, Chung CK, Yun TJ, et al. Epidemiological survey of ossification of the posterior longitudinal ligament in an adult Korean population: three-dimensional computed tomographic observation of 3,240 cases. *Calcif Tissue Int* 2014;94:613–20.
- [12] Wang H, Zou F, Jiang J, et al. Analysis of radiography findings of ossification of nuchal ligament of cervical spine in patients with cervical spondylosis. *Spine (Phila Pa 1976)* 2014;39:E7–11.
- [13] Lee SE, Jahng TA, Kim HJ. Surgical outcomes of the ossification of the posterior longitudinal ligament according to the involvement of the c2 segment. *World Neurosurg* 2016;90:51–7.
- [14] Murayama K, Inoue S, Tachibana T, et al. Ossified posterior longitudinal ligament with massive ossification of the anterior longitudinal ligament causing dysphagia in a diffuse idiopathic skeletal hyperostosis patient. *Medicine (Baltimore)* 2015;94:e1295.
- [15] The Investigation Committee on OPLL of the Japanese Ministry of Public Health and Welfare. The ossification of the posterior longitudinal ligament of the spine (OPLL). *Nihon Seikeigeka Gakkai Zasshi* 1981;55:425–40.
- [16] Kubota M, Baba I, Sumida T. Myelopathy due to ossification of the ligamentum flavum of the cervical spine. A report of two cases. *Spine (Phila Pa 1976)* 1981;6:553–9.
- [17] Matsuzaki H, Hoshino M, Kiuchi T, et al. Dome-like expansive laminoplasty for the second cervical vertebra. *Spine (Phila Pa 1976)* 1989;14:1198–203.
- [18] Chiba H, Annen S, Shimada T, et al. Atlantoaxial subluxation complicated by diffuse idiopathic skeletal hyperostosis. A case report. *Spine (Phila Pa 1976)* 1992;17:1414–7.
- [19] Takasita M, Matsumoto H, Uchinou S, et al. Atlantoaxial subluxation associated with ossification of posterior longitudinal ligament of the cervical spine. *Spine (Phila Pa 1976)* 2000;25:2133–6.
- [20] Shirado O, Azuma H, Takeda N, et al. Quadriplegia complicating atlantoaxial subluxation and ossification of the posterior longitudinal ligament in a patient with rheumatoid arthritis. A case report. *J Bone Joint Surg Am* 2005;87:1354–7.
- [21] Kawabori M, Hida K, Akino M, et al. Cervical myelopathy by C1 posterior tubercle impingement in a patient with DISH. *Spine (Phila Pa 1976)* 2009;34:E709–11.
- [22] Kotani Y, Takahata M, Abumi K, et al. Cervical myelopathy resulting from combined ossification of the ligamentum flavum and posterior longitudinal ligament: report of two cases and literature review. *Spine J* 2013;13:e1–6.
- [23] Shiba M, Mizuno M, Kuraishi K, et al. Cervical ossification of posterior longitudinal ligament in x-linked hypophosphatemic rickets revealing homogeneously increased vertebral bone density. *Asian Spine J* 2015;9:106–9.
- [24] Hirao Y, Chikuda H, Oshima Y, et al. Extensive ossification of the paraspinal ligaments in a patient with vitamin D-resistant rickets: case report with literature review. *Int J Surg Case Rep* 2016;27:125–8.
- [25] Nakashima H, Tetreault L, Nagoshi N, et al. Comparison of outcomes of surgical treatment for ossification of the posterior longitudinal ligament versus other forms of degenerative cervical myelopathy: results from the prospective, multicenter AOSpine CSM-International Study of 479 patients. *J Bone Joint Surg Am* 2016;98:370–8.
- [26] Liu K, Shi J, Jia L, et al. Surgical technique: hemilaminectomy and unilateral lateral mass fixation for cervical ossification of the posterior longitudinal ligament. *Clin Orthop Relat Res* 2013;471:2219–24.
- [27] Feng F, Ruan W, Liu Z, et al. Anterior versus posterior approach for the treatment of cervical compressive myelopathy due to ossification of the posterior longitudinal ligament: a systematic review and meta-analysis. *Int J Surg* 2016;27:26–33.
- [28] Wang T, Tian XM, Liu SK, et al. Prevalence of complications after surgery in treatment for cervical compressive myelopathy: a meta-analysis for last decade. *Medicine (Baltimore)* 2017;96:e6421.
- [29] Liu FY, Yang DL, Huang WZ, et al. Risk factors for dysphagia after anterior cervical spine surgery: a meta-analysis. *Medicine (Baltimore)* 2017;96:e6267.
- [30] Hirabayashi K, Toyama Y, Chiba K. Expansive laminoplasty for myelopathy in ossification of the longitudinal ligament. *Clin Orthop Relat Res* 1999;359:35–48.
- [31] Liu X, Min S, Zhang H, et al. Anterior corpectomy versus posterior laminoplasty for multilevel cervical myelopathy: a systematic review and meta-analysis. *Eur Spine J* 2014;23:362–72.