

## Original Article

# Is atlantoaxial instability the cause of “high” cervical ossified posterior longitudinal ligament? Analysis on the basis of surgical treatment of seven patients

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## Abstract

**Background:** Multilevel ossified posterior longitudinal ligaments (OPLLs), particularly those that extend into the high cervical region, are formidable and challenging surgical problems. The aim of the presentation is to analyze the results of surgical treatment of seven consecutive patients having high cervical OPLL with atlantoaxial and subaxial facet fixations. **Objectives:** We analyze the role of atlantoaxial instability in the management of OPLL that extended into the high cervical region, above the lower border of C3 vertebra. **Materials and Methods:** All patients in the series were males. The age of the patients ranged 48-65 years. Clinical evaluation was done by a 5-point clinical grading scale described by us, Japanese Orthopedic Association (JOA) score, and visual analog scale (VAS). All patients were identified to have relatively “subtle” but definite atlantoaxial facet instability on sagittal imaging and the instability was confirmed by direct handling of the facets during surgery. All patients were treated by multilevel facet fixation that included fixation of atlantoaxial facets. The aim of surgery was stabilization and arthrodesis of the involved spinal segments, as instability was considered to be the prime pathogenetic factor of OPLL. Spinal canal decompression, either by anterior corpectomy or discectomy or by posterior laminectomy or laminoplasty was not done and no attempts were made to remove the OPLL. At an average follow-up of 8 months, all patients showed progressive symptomatic recovery. **Conclusion:** Atlantoaxial facet instability can be a cause or an association of high cervical OPLL. Stabilization of the atlantoaxial joint forms a remarkably effective method of treatment.

**Key words:** Atlantoaxial dislocation, atlantoaxial fixation, ossified posterior longitudinal ligament (OPLL), transarticular facet fixation

## INTRODUCTION

Surgical treatment of ossified posterior longitudinal ligament (OPLL) is riddled with confusion and debate. A number of

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possible etiological factors have been incriminated that possibly initiate the formation and growth of OPLL and maturation of clinical symptoms.<sup>[1-7]</sup> A number of treatment strategies have been suggested.<sup>[8-17]</sup> The vertical, cross-sectional, and horizontal extents of OPLL have implications in surgical decision-making. While a successful clinical outcome may be encouraging, any complication can be devastating for the patient and the family. Despite the number of proposed forms of treatment, there is no ideal or “gold standard” treatment that has been universally accepted and results in uniformly satisfying results. We recently proposed an alternative form of surgical treatment of OPLL that involved only multisegmental spinal fixation as the mode of treatment.<sup>[18,19]</sup> This form of treatment was deployed as spinal instability and was identified to be a major or primary factor responsible for the generation of OPLL. In this presentation, we identify and analyze the role of atlantoaxial instability in the management of OPLL.

### MATERIALS AND METHODS

During the period of November 2014 to August 2015, we treated seven patients having cervical OPLL that extended in the high cervical region, up to or above the lower border of C3 vertebra on sagittal magnetic resonance imaging (MRI) imaging. These patients were analyzed retrospectively. All patients were males and their ages were 48 years, 50 years, 53 years, 55 years, 55 years, 63 years, and 65 years. All the patients had progressive neurological symptoms. The presenting clinical symptoms, radiological features, and outcomes are enumerated in Tables 1-4. A 5-point

clinical grading scale discussed by us recently to assess disability related to craniovertebral junction region myelopathy,<sup>[18,20]</sup> the Japanese Orthopedic Association (JOA) score,<sup>[21]</sup> and visual analog scale (VAS)<sup>[22]</sup> were used to evaluate the patients. Dynamic (flexion and extension views) plain radiography, computed tomography (CT) scan and MRI were done before and after surgery and at follow-up [Figures 1 and 2]. The type of OPLL as assessed on imaging is specified in Table 1. All patients had characteristic multilevel OPLL that resulted in varying degrees of cord compression that led to crippling symptoms and severe neurological deficits. The rostral extent of the OPLL extended at least up to the lower border of C3 vertebra. As per our recently described classification,<sup>[22-24]</sup> we identified that there was Type B facet instability in all cases, meaning thereby that on sagittal imaging the facet of the atlas was dislocated posterior to the facet of axis. It was not possible to identify radiological instability at any other facets joint in the subaxial spine. But all joints were functional and active as could be appreciated from the radiological evidences of intact articular cavity and smooth articular cartilage.

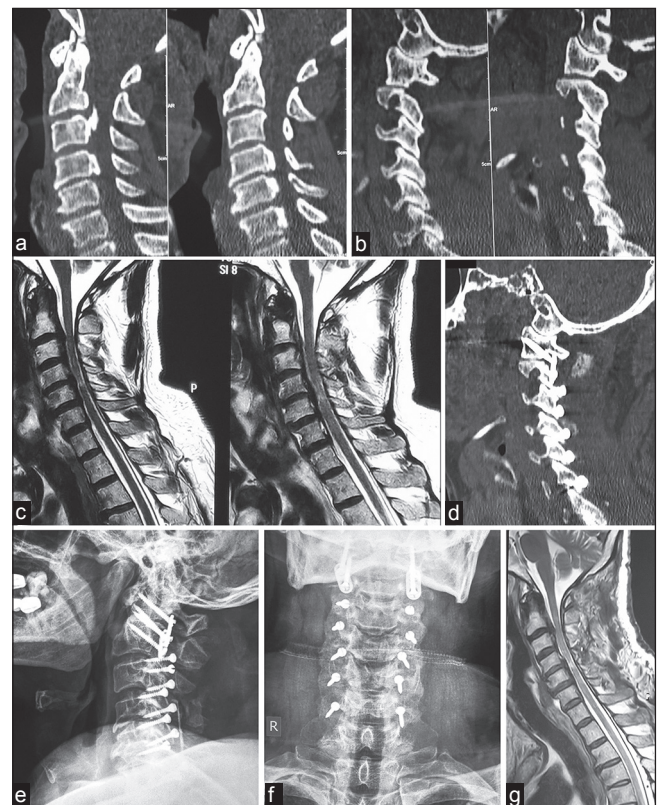
### Operative technique

The patient was placed in the standard prone position with the head held in cervical Gardner Wells traction that kept the head

**Table 1: Table showing the demographics of the patients, types of OPLL, and number of spinal levels affected**

Sex	Number of patients
Male	7
Mean age (years)	55.5 (48-65)
Levels involved	
C1-2	—
C2-3	7
C3-4	7
C4-5	6
C5-6	3
C6-7	1
Number of levels fixed	
C1-C2	7
C2-C3	7
C3-C4	7
C4-C5	6
C5-C6	5
C6-C7	1
Type of OPLL	
Continuous	5
Mixed	2
Segmental	—
Unclassified	—

†OPLL: Ossified posterior longitudinal ligament



**Figure 1: Images of a 65-year-old male patient. (a) Sagittal images of CT scan shows multilevel OPLL (b) Sagittal images through the facets show Type B facet instability (c) T2-weighted MRI shows the cord compression and alteration in the cord signal (d) Postoperative CT scan image showing fixation implant (e) Lateral plain radiograph showing the implant (f) AP view of plain radiograph showing the implant (g) Postoperative T2-weighted image showing the persistence of cord compression and altered cord signal**

**Table 2: Distribution as per clinical grading system**

Grades	Description	Number of patients (preoperative)	Number of patients (postoperative)
Grade 1	Independent and normally functioning	—	2
Grade 2	Walks on own but needs support/help to carry out routine household activities	1	4
Grade 3	Walks with minimal support and requires help to carry out household activities	4	
Grade 4	Walks with heavy support and unable to carry out household activities	1	1
Grade 5	Unable to walk and dependent for all activities	1	—

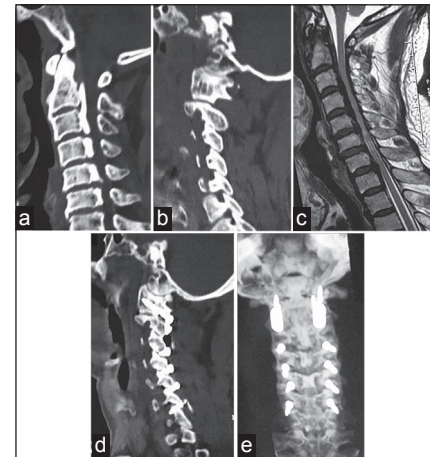
**Table 3: Table showing the preoperative and postoperative clinical assessments as per JOA scoring system**

JOA score	Preoperative (No. of patients)	Postoperative (No. of patients)
<7	1	
8-12	5	1
13-15	1	4
16-17	—	2

**Table 4: Visual analog scale (0 — no pain, 10 — maximum pain)**

VAS score	Preoperative	Postoperative	Postoperative (3 months)
Neck pain	7.5 (4-9)	2 (1-3)	0.5 (0-1)

in a “floating” position. The surgical position was adopted from our earlier described position used for surgery for atlantoaxial fixation.<sup>[25-28]</sup> Traction was deployed essentially to keep the head and neck stable during surgery and the face off the head holder, avoiding any pressure injury to the eyes and facial skin. Midline incision was taken and employing subperiosteal dissection, the articular joints and facets of the cervical spine were exposed widely on both sides. The spinal segments that were exposed depended on the number of cervical levels involved by OPLL. Apart from treating the levels of cervical spine involved by OPLL, direct observation of the facet articular and manual manipulation were used to assess the levels of spinal instability. Atlantoaxial fixation was done first and subsequently subaxial spinal fixation was carried out, essentially by similar surgical strategy. The atlantoaxial joint was widely exposed. C2 ganglion was resected on both sides in one patient to widely expose the atlantoaxial joint. In other cases, the C2 ganglion was elevated superiorly. The articular capsule of the C1-2 joint and subaxial joints was sharply cut and the joints were exposed. Varying sizes of osteotomes were used and were introduced with their flat, sharp edge and then turned 90° in a screwing motion to affect denuding of the articular capsule and cartilage endplate. Bone graft pieces harvested from the iliac crest were jammed into the articular cavity. C1-2 facet fixation was done employing plate and screw technique described by us earlier in 1988.<sup>[28]</sup> The titanium material screws used for the fixation of facets of both atlas and axis measured 2.8 mm in thickness and ranged



**Figure 2: Images of a 55-year-old male. (a) Sagittal image of CT scan showing multisegmental OPLL (b) Sagittal image of CT scan with the cut through the facets showing Type B facet instability (c) T2-weighted MRI showing multilevel cord compression by the OPLL (d) Postoperative CT scan showing the multilevel spinal facet fixation (e) Anteroposterior view of the CT scan showing the fixation**

26-28 mm in length. Transarticular facet fixation by the technique described earlier by Roy-Camille<sup>[29]</sup> was deployed for subaxial spinal stabilization. For the transarticular method of fixation, the screws used were of 14 mm length and 2.8 mm in diameter. The interspinous process and interlaminar ligaments were then widely removed. The spinous process is sectioned along its base. Bone graft obtained following sectioning of the adjoining spinous processes and that obtained from the iliac crest was placed over the cut surface of the base of the spinous process and the host bone area of the laminae was appropriately prepared by drilling its outer cortex. Neurophysiological monitoring was not used during surgery. The patient was then advised limited cervical movement using external orthosis for a period of 8 weeks. After this period and confirmation of preliminary evidences of spinal fusion (that included a stable and satisfactory screw positioning on dynamic imaging), all routine activities were permitted.

**RESULTS**

The follow-up period ranged 3-12 months, the average being 8 months. The clinical outcome of surgery is elaborated in Tables 2-4. The clinical evaluation included the location and degree of preoperative and postoperative pain and myelopathy symptoms using an analog scale, subjective and objective



alterations in sensation and weakness, walking difficulty, and bowel or bladder changes. All patients improved in symptoms in the “immediate” postoperative period following surgery. During the period of follow-up, the improvement in neurological symptoms persisted [Tables 2-4]. There has been no need for any additional surgical maneuver in any patient. All patients underwent evaluation on static and dynamic cervical spine radiographs and CT scanning and MRI. Static neutral lateral radiographs were used to assess cervical sagittal balance, whereas anteroposterior radiographs were used to exclude preoperative abnormal coronal alignment [Figure 1]. All patients had loss of cervical lordosis and straightening of the cervical spine before surgery that persisted after the operation. The lordotic angle was measured using Cobb's method of measurement and there was no significant difference between the preoperative and postoperative values. There was no implant-related complication. Successful bone fusion was observed in all cases. Fusion of the spinal segment was defined as an absence of all kinds of motion and alterations in the interlaminar and intervertebral body distances on flexion-extension radiographs. Bone formation was additionally observed inside and across the facets and over the laminae. On follow-up imaging, the dimensions of OPLL regressed or increased in none of the patients.

## DISCUSSION

OPLL is a relatively rare but well-described and elaborately discussed clinical entity. Although identified throughout the world, the number of scientific articles from Asia outnumbered papers from other continents. Dietary, environmental, and physical body constitution-related factors, apart from a host of other possible etiological factors, have been suggested to be the possible causes.<sup>[5-7]</sup> In general, the patients harboring OPLL are marginally or significantly obese and have a relatively sedate lifestyle. The devastating nature of complications of surgery makes the treatment of patients having OPLL one of the most feared surgical issues. The surgical treatment is difficult to conceptualize essentially because the exact pathogenesis of the abnormal bone formation is unclear.

OPLL that extends in the high cervical region is particularly more formidable to surgical treatment. Anterior C2 corpectomy, removal of the arch of the atlas and odontoid process, and decompression of the cord by removal of OPLL in the craniovertebral junction have only rarely been advocated.<sup>[30,31]</sup> In general, a majority of the surgeons dealing with lesions at this location have advocated laminectomy or laminoplasty for indirect decompression of the spinal cord in an attempt to increase the volume of the spinal canal and accommodate the additional and uninvited intrusion. There have been a number of reports in the literature that indicate the need of a wide removal of the OPLL as it has been observed that any treatment that does not remove the primary cause of compression can be wrought with failure and does not stall the progressive and relentless growth of the OPLL. Any surgical treatment that involves

removal of the high cervical and craniovertebral junction OPLLs is not only technically difficult but a wide exposure and radical removal of OPLL is only rarely possible and has never been demonstrated by imaging. Moreover, the surgical complications can be unforgiving. The search of an ideal surgical treatment of long and multisegmental OPLLs, particularly those that extend into the high cervical region continue.

OPLL is conventionally associated with a stable or more than normally stable disorder. Bone formation along the ligaments is considered to be an additionally stabilizing factor. As the lesion is considered to be “stable,” the treatment involves decompression of the cord, removal of the OPLL, or widening of the spinal canal size by removing bone anterior, posterior, or circumferentially around the cord. The surgical procedures that involve stabilization are aimed to prevent the destabilizing effects of the “decompressing” bone removal surgery. The twin operative strategy that involves decompression and fixation, either by anterior or by posterior surgical route, has been generally adopted. However, despite the surgical experience and expertise of the surgeon, such a surgical procedure and the maneuver of resection of OPLLs, particularly those that extend into high cervical region, can be tedious, time-consuming, and potentially hazardous. We had recommended oblique corpectomies for exposure and resection of the OPLL and had suggested that such a conservative bone removal procedure does not affect the stability of the spine and additional stabilization procedures can be avoided.<sup>[32,33]</sup>

Craniovertebral junction instability has been identified to be associated with secondary effects on the cervical spine and even in the rest of the spine. Short neck, torticollis, and bone fusion have been associated with craniovertebral junction instability.<sup>[20,33,34]</sup> We recently identified the association of facet instability with longstanding or relentlessly progressive pathologic phenomenon at the craniovertebral junction of Chiari 1 malformation and syringomyelia. We proposed that Chiari 1 malformation could simulate Nature's air bag that is positioned in place to provide a cushion for critical neural structures and avoid their pinching between bones in the event of potential or manifest instability.<sup>[20,33]</sup> We also identified that facet instability can be associated with Group B basilar invagination. In this group of basilar invagination, there are evidences of longstanding and progressive pathological phenomenon. We recently identified an association of atlantoaxial facet instability in cases with multilevel cervical spondylotic disease and recommended atlantoaxial fixation in such cases.<sup>[24]</sup> Our literature search did not reveal any article that related craniovertebral instability to cervical OPLL. Conventionally, instability at the craniovertebral junction has been identified by alteration in the atlantodental interval. We recently identified facet malalignment as an important measure of instability at the craniovertebral junction.<sup>[23]</sup> Facet instability may or may not be associated with odontoid process-related cord compression or symptoms. As cord compression is not a prominent or major issue, the symptoms related to facet instability can be subtle and longstanding. Although there is no

morphological study that evaluates the relationship of the facets of the atlas and axis during dynamic movements, it does seem that some degree of facet malalignment can be within the range of normal variation. In the presented series of patients, we identified the presence of Type B facet instability in all cases. Apart from instability observed on imaging, direct intraoperative observations confirmed the presence of instability.

The role of spinal instability in the generation and growth of OPLL has been only been infrequently discussed in the literature. We recently hypothesized and analyzed the role of instability in the management of OPLL. We identified that stabilization of the spinal segments alone without any form of bone or soft tissue decompression can have a defining role in the treatment of OPLL.<sup>[18]</sup> Identification of subaxial facet instability on radiological imaging is difficult due to their oblique profile. However, facet instability at the atlantoaxial joint can be identified relatively easily due to their more horizontal lie and rectangular box-like alignment. On the basis of our 30-year experience in dealing with the atlantoaxial instability and with direct handling of the facets of the atlas and axis, we realize that the atlantoaxial joint in cases with OPLL that extends into the high cervical region is unstable. The instability is relatively subtle but remarkable. This fact is also suggested by the presence of facet malalignment on imaging and by the presence of an active and functioning joint with well-defined articular surface despite the multilevel presence of additional abnormal bone of OPLL. Additionally, we observed instability of spinal segments involved by OPLL by direct observation of the status of articular cavity and facets and direct manual handling and manipulations. We used our recently described clinical grading scale<sup>[20]</sup> for evaluation of the patient. However, as the scale is not validated as yet, we additionally used the standard and recognized JOA score. Our satisfactory clinical outcome also reinforces our proposed concept that atlantoaxial instability may have a defining role in the generation and progression of OPLLs in general and particularly those that extend into the high cervical region. It also appears that facet malalignment on imaging may not be the only criterion to detect atlantoaxial instability. In Type C facet instability, the instability of the region is detected only by direct handling of the facets. We label such instability as “central” or “axial” instability.<sup>[18,23]</sup> Although the procedure of atlantoaxial fixation is technically demanding, it is relatively simpler when compared to other possible surgical options in such cases. Our surgical outcome analysis suggests that missing or ignoring the presence of atlantoaxial facet instability can be an important cause of suboptimal result or failure of surgery for high cervical OPLL.

In all our seven cases, atlantoaxial fixation was done in addition to other cervical level fixation. From the study, it is unclear if fixation of other spinal segment could have been avoided. The contribution of atlantoaxial dislocation in the overall spinal instability is also difficult to assess. The number of cases treated in the series is relatively small. The relative rarity of such cases makes the adoption of appropriate scientific protocol difficult. The negative aspect of the study is that the surgical decision-

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making had a subjective influence that was based on the personal operative experience of manual handling of facets of the spine. Although the total study period and duration of follow-up are relatively small, the remarkable clinical recovery following the treatment has prompted the reporting of observations.

## CONCLUSION

It appears that multilevel spinal stabilization may have a leading role to play in the treatment of OPLL. Atlantoaxial fixation may be done in all cases where the OPLL extends into the high cervical region, up to or superior to the inferior border of C3. Atlantoaxial fixation should be done irrespective of the presence or absence of facet malalignment on imaging.

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## Conflicts of interest

There are no conflicts of interest.

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