New Development and Progression of Ossification of the Posterior Longitudinal Ligament (OPLL) after Cervical Disc Arthroplasty: A Case Report

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Abstract

Purpose: To report an extremely rare case of new development and progression of ossification of posterior longitudinal ligament (OPLL) after cervical disc arthroplasty (CDA).

Background: To our knowledge, new development and progression of ossification of OPLL after CDA have not been reported.

Methods: A 44-year-old female with cervical disc herniation at C5-6 presented with radiculopathy. The patient had no evidence of preexisting OPLL on plain radiographs and magnetic resonance imaging. She underwent CDR at C5-6 and her symptoms were significantly improved after surgery.

Results: New development of OPLL at C5-7-T1 was identified for the first time at 6 years after CDA at C5-6, and progression of OPLL was observed at 8 and 13 years' follow-up. Due to new development and progression of OPLL, there was no segmental motion at C5-6 with CDA. The patient continued to follow up without further surgery because there was no deterioration in clinical symptoms.

Conclusion: Our study demonstrated an extremely rare case of new development and progression of OPLL after CDA with a long term follow-up. Our case suggests potential clues for discovering the complex pathological mechanism of OPLL.

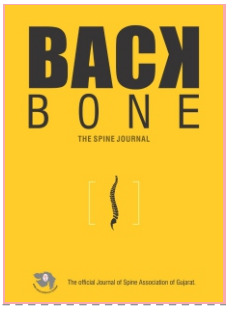
Keywords: Ossification of the posterior longitudinal ligament, New development, Progression, Cervical disc arthroplasty.

Introduction

Ossification of the posterior longitudinal ligament (OPLL), first described in 1838, is a condition characterized by progressive abnormal ossification growth of the posterior longitudinal ligament (PLL). OPLL can result in significant spinal canal compromise with cord compression and neurological injury [1-3]. Surgical intervention should be considered for patients with neurologic symptoms such as myelopathy and/or radiculopathy [4]. A variety of surgical approaches have been described utilizing anterior, posterior, and combined anteroposterior approaches, each with unique risks and benefits [5].

Many studies have reported on the pathogenesis of the progression of preexisting OPLL after cervical spine surgeries [6-11]. The rate of postoperative progression of OPLL is higher in posterior-approach surgeries than in anterior-approach surgeries. When considering only posterior-approach surgeries, motion-preserving laminoplasty has shown a higher rate of OPLL progression than the laminectomy and fusion procedure [12-18].

There are very few reports on progression of OPLL after anterior cervical spine surgeries [9,11]. To our knowledge, however, new development and progression of ossification of posterior longitudinal ligament (OPLL) after cervical disc arthroplasty (CDA) have not been reported. Therefore, we report an extremely rare case of new development and progression of OPLL with a long term follow-up after CDA surgery.



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Case Report

A 44-year-old female presented with neck pain (neck visual analogue scale [VAS] score: 5) and both shoulder and arm radiating pain (arm VAS score: 7/7) for 3 months. Neurological examination was otherwise within normal limits

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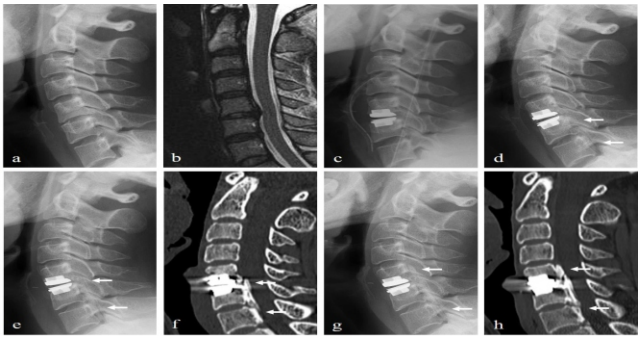

Figure 1: Lateral radiograph (a) and sagittal MRI (b) of the cervical spine showing disc herniation and spondylosis at the C5-6 level in the absence of preexisting OPLL. Patient underwent cervical disc arthroplasty (CDA) at C5-6 using Prodisc-C® ©. Follow-up lateral radiograph (d) at postoperative 6 years showing good maintenance of the CDA at C5-6 and new development of OPLL at C6 and C7 (white arrows). Follow-up lateral radiograph at postoperative 8 years showing further progression of mixed-type OPLL at C5-7-T1 (white arrows) (e). Sagittal reconstructed CT scan at postoperative 8 years showing good maintenance of the CDA at C5-6 and further progression of mixed-type OPLL to C5-6 disc space and T1 (white arrows) (f). Follow-up lateral radiograph at postoperative 13 years showing more longitudinal progression of mixed-type OPLL with the extension of C5 upper body and T1 (white arrows) (g). Sagittal reconstructed CT scan at postoperative 13 years showing good maintenance of the CDA at C5-6 and more longitudinal progression of mixed-type OPLL to the upper margin of the C5 body and T1 (white arrows) (h).

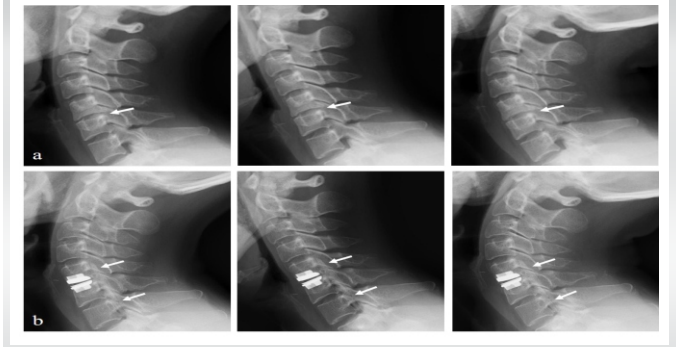

Figure 2: In terms of segmental motion at C5-6, preoperative radiographs showed little segmental motion (white arrows) due to severe spondylosis and loss of disc height more than 50% (Figure 2a). However, due to longitudinal progression of OPLL to C5 upper body and C5-6 disc space and T1 (white arrows), there was no segmental motion at C5-6 with the cervical disc arthroplasty (CDA) at 13 years after CDA (Figures 2b).

without pathologic reflex. Lateral radiograph (Figure 1a) and sagittal magnetic resonance imaging (MRI) (Figure 1b) of the cervical spine showed disc herniation and spondylosis at the C5-6 level in the absence of preexisting OPLL. She underwent anterior cervical discectomy, resection of posterior longitudinal ligament, uncoforaminotmy, and CDA at C5-6 using Prodisc-C® (DePuy Synthes, Paoli, PA, USA) (Figure 1c). Her symptoms were significantly improved after surgery, so she did not require any immediate outpatient clinic visits. However, 6 years after surgery, she visited an outpatient clinic complaining of stiff and uncomfortable neck (neck VAS score: 3) but improved neurologic outcomes were well maintained without deterioration. Follow-up lateral radiograph of the cervical spine revealed good maintenance of the CDA at C5-6 and new development of OPLL at C6 and C7 (Figure 1d). Eight years postoperative, follow-up radiograph (Figure 1e) and sagittal reconstructed computed tomography (CT) scan (Figure 1f) showed good maintenance of the CDA at C5-6 and further progression of mixed-type OPLL to C5-6 disc space and T1. Thirteen years postoperative, follow-up radiograph (Figure 1g) and sagittal reconstructed CT scan (Figure 1h) showed good maintenance of the CDA at C5-6 and more longitudinal progression of mixed-type OPLL to the upper margin of the C5 body and T1.

In terms of segmental motion at C5-6, preoperative radiographs showed little segmental motion due to severe spondylosis and loss of disc height more than 50% (Figure 2a). However, due to longitudinal progression of OPLL to C5 upper body and C5-6 disc space, there was no segmental motion at C5-6 with CDA at 13 years after CDA (Figures 2b). She continued to follow up without further surgery because there was no deterioration in clinical symptoms.

Discussion

While the pathophysiology of OPLL remains poorly understood, multiple genetic, biologic, and mechanical factors have been related to new development and progression of OPLL [6-8]. Recent studies revealed the mechanisms of the OPLL as follows: (1) direct stimulation of PLL by surgical procedure, (2) mechanical stress induced by postoperative structural changes, (3) postoperative instability after surgery, and (4) spontaneous increase in ossification itself [6-8, 17-19]. Especially, posterior instrumented fusion has the effect of reducing the OPLL growth rate by preventing the mechanical stress compared with motion-preserving laminoplasty, however, it does not necessarily mean that OPLL progression can be completely prevented. Similar to the preventive effect of OPLL in posterior instrumented fusion, in the case of anterior cervical discectomy and fusion, the occurrence or progression of OPLL can be reduced by preventing the mechanical stress. However, there are very few reports on progression of OPLL after anterior cervical spine surgeries, especially C-ADR and the etiology of OPLL and its mechanism of development remains unclear and has not been fully identified. Therefore, we would like to report an extremely rare case of new development and progression of OPLL with a long-term follow-up after CDA surgery.

Numerous studies have reported on OPLL progression and established risk factors for progression, including types of OPLL, timing and location of OPLL progression, and type of surgeries [9-18]. When considering the impact of OPLL type on the progression of OPLL, it should be noted that OPLL type could change during the follow-up period [10-12]. Chiba et al. reported a higher rate of postoperative OPLL progression in continuous-type and mixed-type OPLL [20]. Similarly, in our case, the mixed-type OPLL progressed with more longitudinal extension to the upper margin of the C5 body. At 1 year after CDA, there was no evidence of new formation of OPLL. However, new formation of OPLL was first detected at C6 and

C7 at postoperative 6 years after CDA, and gradually progressed to C5 upper body and T1 at postoperative 8 and 13 years after CDA. In terms of segmental motion at C5-6, there was little segmental motion at C5-6 at preoperative radiographs including flexion and extension due to severe spondylosis, loss of disc height more than 50% and uncovertebral osteophytes. At 1 year after CDA, little segmental motion at C5-6 was found, especially in extension, with no evidence of OPLL formation. At 6 years after CDA, little segmental motion at C5-6 was still found, especially in extension, with new formation of OPLL at C6 and C7. However, due to longitudinal progression of OPLL to C5 upper body, C5-6 disc space and T1, there was no segmental motion at C5-6 with CDA at 8 and 13 years after CDA.

The timing and process of progression are important for a better understanding of OPLL progression [12]. Progression of OPLL was more likely to occur in the early phase after surgery, and the growth rate decreased over time [21]. Therefore, follow-up in the early stages after operation was important in evaluating progression of OPLL [12]. Chiba et al conducted the first multicenter study to investigate the incidence of OPLL progression after posterior decompression and determined that the incidence of postoperative progression was 38.9% at 1 year and 56.5% at 2 years after surgery, thereby concluding that most progression occurred within the first 2 years after surgery [20]. In our study, the timing of OPLL development was within the first 6 years postoperative. However, it is difficult to know the exact timing of the new development of OPLL because the patient had an excellent postoperative recovery and did not visit the hospital for six years due to lack of relapse of cervical symptoms. Therefore, the new development of OPLL was identified for the first time at six years postoperative. Furthermore, the symptoms did not recur after that, so the only follow-up was observed at 8 and 13 years postoperative.

Regarding the location of OPLL progression, the most common location of OPLL in the cervical spine is C4-5 [22], and OPLL progression was frequently observed at C2, C3, and C4 and marked at the upper cervical level rather than at the lower levels.[12] Interestingly, our case with C-ADR showed development of OPLL to the distal C5-T1, including the C-ADR site, and simultaneously more progression of the proximal extension of the C5 body.

OPLL progresses faster in surgically managed patients than in conservatively managed patients; for example, the development of OPLL was found to be much more rapid in patients who underwent surgery (laminectomy or laminoplasty) compared with a nonsurgical cohort [13, 23]. Specifically, surgery type is a significant factor in postoperative OPLL progression [20]. Lee et al reported that posterior instrumented fusion has the effect of reducing the OPLL growth rate compared with motion-preserving laminoplasty [16]. The prevalence of radiological OPLL progression in the laminoplasty group (62.5%) was significantly higher when compared with the fusion group (7.6%) [17]. In addition, possible mechanisms of new development and progression of OPLL may closely be related with the hypermobility by CDA or excessive removal of uncovertebral joint and posterior longitudinal ligament by decompression. Several previous papers reported that severe spondylosis is one of the most important risk factors for heterotopic ossification formation after CDA [24, 25]. Similar to heterotopic ossification formation after CDA, I think that preexisting severe spondylosis may provocate new formation and progression of OPLL after CDA in this case. In addition, inappropriate resection of posterior longitudinal ligament may be also involved in new formation and progression of OPLL.

Additionally, long-term follow-up of patients after laminoplasty has demonstrated radiographic progression of OPLL, though neurologic deterioration and reoperation for symptomatic progression is rare [15]. Therefore, prophylactic surgery for asymptomatic patients is not routinely recommended; patients with severely stenotic cervical segments and cord signal changes should instead be closely monitored and questioned for signs or symptoms of myelopathy because progression of OPLL does not positively correspond with neurologic deterioration [26, 27]. Similarly, despite the new development and progression of OPLL, the patient in our study showed any neurological deterioration or required further surgery.

While multiple genetic and environmental factors have been related to the development of OPLL, there was no history of genetic and environmental factors including diabetes mellitus, obesity, hypothyroidism, and a high sodium diet in our two patients. However, we did not evaluate the genetic components as evidenced thorough familiar inheritance and genetic analysis such as BMP4, BMP9, and COL6A1.

Conclusion

Our study demonstrated the first case of new development and progression of OPLL after CDA with a long-term follow-up. Our case suggests potential clues for discovering the complex pathological mechanism of OPLL.

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Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his/her consent for his/her images and other clinical information to be reported in the Journal. The patient understands that his/her name and initials will not be published, and due efforts will be made to conceal his/her identity, but anonymity cannot be guaranteed.

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