



## Review Article

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# Ossification of the Posterior Longitudinal Ligament: Surgical Approaches and Associated Complications

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Ossification of the posterior longitudinal ligament (OPLL) is a rare but potentially devastating cause of degenerative cervical myelopathy (DCM). Decompressive surgery is the standard of care for OPLL and can be achieved through anterior, posterior, or combined approaches to the cervical spine. Surgical correction of OPLL via any approach is associated with higher rates of complications and the presence of OPLL is considered a significant risk factor for perioperative complications in DCM surgeries. Potential complications include dural tear (DT) and subsequent cerebrospinal fluid leak, C5 palsy, hematoma, hardware failure, surgical site infections, and other neurological deficits. Anterior approaches are technically more demanding and associated with higher rates of DT but offer greater access to ventral OPLL pathology. Posterior approaches are associated with lower rates of complications but may allow for continued disease progression. Therefore, the decision to pursue either an anterior or posterior approach to surgical decompression may be critically influenced by complications associated with each procedure. The authors critically review anterior and posterior approaches to surgical decompression of OPLL with particular focus on the complications associated with each approach. We also review the recent work in developing new surgical treatments for OPLL that aim to reduce complication incidence.

**Keywords:** Postoperative complications, Neurosurgical procedures, Cervical vertebrae, Spinal diseases, Ossification of posterior longitudinal ligament



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## INTRODUCTION

Pathologic ossification of the posterior longitudinal ligament (OPLL) is a disease that is a relatively uncommon, but nonetheless important, cause of cervical myelopathy. Untreated, it may lead to neurologic morbidity and ultimately significant loss of independence.<sup>1</sup>

The incidence of OPLL in the general population varies. It is significantly influenced by geographic location and ethnicity, with the highest reported prevalence in East Asian countries.

The 12-year prevalence in Korea was found to be 2.04%, while studies of the prevalence in Japan range between 1.9%–4.3% of all patients with cervical spine disorders.<sup>1,2</sup> In North America, a single institution reported the incidence of OPLL to be 2.2% (95% confidence interval [CI], 1.7%–2.8%) with the highest incidence in Asian American subpopulations ( $p=0.005$ ).

Typically the average age of symptom onset falls between 50–65 years,<sup>2–5</sup> with some studies in Japanese populations reporting the average onset to be as early as 51 years in men and 49 years in women.<sup>1</sup> Recently, a Korean national database study found

the peak age of annual incidence to be between 60–75 years, much later than previously reported.<sup>2</sup> Interestingly, this study found a slight predominance of women being affected by OPLL, which is in relative contrast to the bulk of the literature, which hitherto reported a 2:1 male to female predominance.<sup>1,2,4,5</sup> In the Korean study, men did, however, comprise a significantly larger proportion of those undergoing surgical correction of OPLL (60.99% vs. 39.01%,  $p < 0.001$ ).<sup>2</sup>

### 1. Pathophysiology of OPLL

The posterior longitudinal ligament (PLL) is located within the spinal canal, along the dorsal surface of the vertebral bodies. It originates at the axis and extends distally to the sacrum. The PLL functions primarily to resist hyperflexion of the spine, and normally consists histologically of flat, well-aligned fibroblasts with no intervening cartilaginous elements.<sup>6</sup> Pathologic OPLL is attributed to the proliferation and differentiation of fibroblast-like chondrocytes and osteoblasts, concurrent with neovascularization of the ligament which may be caused by overexpression of bone morphogenetic proteins (BMPs) in the ossified ligament.<sup>7</sup> This results in the normally well-organized fibers comprising the native ligament being replaced with cartilaginous tissues, as well as centers of hyalinoid degeneration and ossification.<sup>6</sup>

Genetic risk factors contribute significantly to the development of OPLL. In one study of 347 families, OPLL was incidentally discovered in up to 30% of affected patients' siblings.<sup>8</sup> The genes implicated in OPLL, such as COL6A1, BMP, transforming growth factor-beta, and others, are critical mediators of collagen integrity and bone homeostasis.<sup>9–11</sup> OPLL has also been linked to other disorders of aberrant ossification and bone homeostasis, such as diffuse idiopathic skeletal hyperostosis.<sup>12</sup>

### 2. Presentation

OPLL can affect any spinal segment but is most frequently observed in the cervical spine. Reasons for this are not entirely clear, however may be related to the narrower caliber of the cervical spinal canal with the increased motion that may predispose this segment to symptom development. The incidence of concomitant OPLL in other, remote spinal levels, is high; one study noted that 56.2% of patients with cervical OPLL also had evidence of disease in the thoracolumbar spine.<sup>13</sup> While most patients with OPLL will present with chronic progressive cervical myelopathy in the 4th or 5th decade, as many as 15% of patients with OPLL may present with acute symptoms related to trauma.<sup>1</sup> The patient's baseline degree of myelopathy upon ini-

tial presentation appears to play a critical role in predicting the progression of disease and the likelihood of that patient benefiting from surgery. Matsunaga et al.<sup>1</sup> found that 71% of patients who were myelopathy-free at first presentation remained myelopathy-free at 17-year follow-up.

### 3. Evaluation & Classification of Patients

Physical examination is paramount in assessing the patient with OPLL and treatment is based on their degree of neurologic dysfunction. Most classifications therefore are based on myelopathy or cord dysfunction. There are several classification systems currently used to convey severity of disease at presentation. The 2 most ubiquitous are the Nurick scale for classification of myelopathy (adapted in Table 1) and the Japanese Orthopedic Association (JOA) scale for classification of cervical myelopathy. The JOA scale is based on upper and lower extremity motor and sensory function as assessed using a patient's ability to complete certain tasks. The JOA score is often used to report recovery rate using the formula:

$$\text{Recovery rate} = (\text{postoperative JOA score} - \text{preoperative JOA score}) \times 100 / (17 - \text{preoperative JOA score}).^{14}$$

Postoperative recovery rate is thus defined as 100% (complete recovery), more than 75% (excellent), more than 50% (good), more than 25% (fair), more than 0% (unchanged), and less than 0% (poor).

Plain radiography, dynamic imaging, magnetic resonance imaging (MRI), and computed tomography (CT) are all crucial in the work-up of the OPLL patient. The Japanese Ministry of Public Health and Wellness (JPMPhW) classification system of OPLL is based exclusively on disease appearance on lateral radiography, and is the most widely used classification system.<sup>15</sup> Axial CT is paramount though, as it allows for the assessment of disease severity as measured by the degree of canal compromise by the OPLL mass. Foci of OPLL that occupy  $\geq 50\%$  of the spinal canal have repeatedly been associated with more severe preoperative disease, as well as lower rates of postoperative improvement.<sup>1</sup> In 2014, the JPMPhW proposed a second, more

**Table 1.** Nurick classification of myelopathy

Grade 0	No signs or symptoms of cord dysfunction
Grade 1	Normal gait despite exam findings of cord compression
Grade 2	Mild gait impairment that does not prevent employment
Grade 3	Moderate gait impairment that limits employment but does not require assistance
Grade 4	Assistance or assistive devices required for ambulation
Grade 5	Wheelchair- or bed-bound

**Table 2.** Summary of surgical approaches to OPLL and their associated advantages, disadvantages, and complications

Approach	Advantages	Disadvantages	Complications
Anterior ADF, ACDF	Direct decompression of OPLL Improved outcomes in cases of severe stenosis (> 60% occupancy) Reduced progression of OPLL Less extensive tissue dissection and instrumentation	Higher degree of difficulty in access Technically demanding Less preservation of cervical ROM	Dural tear/CSF leak (2.4% <sup>32</sup> –31% <sup>39</sup> ) Dysphagia, dysarthria, hoarseness (2.4% <sup>32</sup> –16.8% <sup>55</sup> )
Posterior LAMP, LF	Technically less demanding Well-suited for broad range of patients regardless of disease severity or comorbidities Less aggressive tissue disruption Lower rates of OPLL progression/recurrence (LF > LAMP) <sup>38</sup> Preserved cervical ROM	Higher rates of OPLL progression Higher rates of postoperative complications Indirect decompression of OPLL	C5 palsy LAMP (9.6%–25%) <sup>33</sup> LF (0%–8%) <sup>35</sup> Axial pain <sup>58</sup> LAMP (22.2%) LF (23.2%)

Key advantages and disadvantages associated with the various approaches to surgical decompression of OPLL and the most common complications encountered with each approach.

OPLL, ossification of the posterior longitudinal ligament; ADF, anterior decompression with floating; ACDF, anterior cervical decompression and fusion; ROM, range of motion; CSF, cerebrospinal fluid; LAMP, laminoplasty; LF, laminectomy and fusion.

detailed classification system based on CT, however this system has not as of yet been fully embraced.<sup>16</sup> In the original JPMHW system, OPLL can be divided into 4 distinct subtypes, based on the degree of segmental involvement:

- (1) Localized, consisting of a solitary OPLL lesion involving an isolated vertebral level;
- (2) Segmental, consisting of multiple, separate vertebral level lesions;
- (3) Continuous, where a single uninterrupted lesion involves multiple interspaces, and;
- (4) Mixed, which is a combination of localized, segmental, and continuous.

The most frequently reported type of OPLL varies, with many studies reporting a higher incidence of the segmental type (type 2)<sup>3,17,18</sup> and others reporting a nearly equal distribution of segmental, continuous, and mixed types.<sup>3,19</sup>

#### 4. Surgical Management

Surgical intervention has consistently been shown to significantly improve clinical outcomes for most patients with moderate to severe, or progressive, myelopathy attributable to OPLL.<sup>20</sup> The degree of clinical improvement achieved with surgery has been found to be similar to the surgical benefits achieved with decompression of other forms of degenerative cervical myelopathy.<sup>21</sup> The degree of benefit achieved with surgery is significantly determined by the degree of myelopathy at presentation. Patients who present with absent or minimal myelopathy (Nurick grade 1 or 2) do not benefit from surgery over those managed conservatively while those with severe myelopathy (Nurick grade 3 or 4) benefit substantially from surgery.<sup>1</sup> The risk for neuro-

logic deficit in patients with minimal myelopathy is still considerable: patients with OPLL managed conservatively were found to have a cumulative incidence rate of hospitalization for spinal cord injury that was significantly higher (2.98%; 95% CI, 1.41%–6.32%) than matched controls (0.19%; 95% CI, 0.08%–0.48%).<sup>22</sup>

The primary goal of surgical management of OPLL is to decompress the neural elements by either (1) direct resection or thinning of the offending mass, or (2) expansion of the spinal canal to accommodate the OPLL lesion. Secondly, surgical intervention aims to restore or otherwise preserve alignment and stability. Surgical intervention can be undertaken from an anterior, posterior, or combined approach. The following discussion will aim to address the advantages, disadvantages, and complications associated with these approaches (summarized in Table 2).

## DISCUSSION

### 1. Anterior Approach

The anterior approach to OPLL can have a higher degree of difficulty since the ossified ligament is typically densely adherent to the ventral dura, manipulation of which has a relatively high incidence of cerebrospinal fluid (CSF) leaks. However, with focal OPLL this technique can be very effective. The anterior approach to OPLL decompression involves standard exposure as described by Smith-Robinson and others.<sup>20,23</sup> The OPLL itself can be resected completely, or otherwise thinned and released in what Yamaura et al.<sup>24</sup> first described as the anterior decompression with floating (ADF).

Anterior approaches to OPLL, while potentially more technically demanding than more typical degenerative pathology, af-

fords the same advantages that are otherwise enjoyed when addressing ventral pathology. Perhaps the most important of these advantages is the direct access. Several studies have demonstrated that while long-term clinical benefit is equivocal among anterior and posterior approaches when the degree of cervical stenosis < 60%, significantly better recovery rates and postoperative JOA scores have been reported with anterior decompression when spinal canal stenosis is greater than 60%.<sup>25,26</sup> A nonrandomized prospective study of 150 patients with OPLL demonstrated that anterior decompression achieved superior JOA scores at last follow-up compared to posterior laminoplasty (LAMP) when spinal canal stenosis exceeded 50% ( $p < 0.05$ ).<sup>27</sup>

Furthermore, anterior approaches are associated with a decreased rate of continued OPLL progression since the OPLL is removed. One study found that nearly 50% of patients treated with posterior LAMP demonstrated progression of disease at 5-year follow-up, compared to only 5.0% of patients treated with ADF ( $p > 0.01$ ).<sup>28</sup> Direct decompression of OPLL may also explain why patients with spinal cord signal changes on MRI tend to have better outcomes with anterior decompression. One randomized, controlled trial (RCT) of 56 patients with OPLL demonstrated that patients with preoperative spinal cord changes on MRI (T1 hypointensity or T2 hyperintensity) experienced a greater degree of improvement following anterior decompression and fusion compared to posterior laminectomy ( $p < 0.05$ ).<sup>29</sup>

Anterior approaches also typically require less extensive tissue dissection and instrumentation, increased recovery time, and better restoration of lordosis. One prospective single institution study of 42 patients treated with either ADF or the so-called “French-door” LAMP demonstrated preserved range of motion (ROM) and C2–7 lordotic angle, and minimal postoperative kyphotic progression, with ADF compared to LAMP at 5-year follow-up.<sup>28</sup> One nonrandomized prospective study of 150 consecutive patients demonstrated no significant difference in JOA or VAS scores of patients treated with anterior decompression or LAMP ( $p < 0.05$ ).<sup>27</sup> In this study, however, anterior decompression was associated with preserved preoperative ROM, while LAMP was associated with a 6.82% decline in ROM at last follow-up ( $p < 0.05$ ).<sup>27</sup>

## 2. Posterior Approach

The posterior approach is the more commonly used procedure for the treatment of OPLL.<sup>21</sup> It is technically less demanding, and can be applied to a broad range of OPLL patients regardless of disease severity or degree of systemic comorbidity. This is reflected in a study by Morishita et al.,<sup>30</sup> which found

that, prior to propensity matching in over 8,000 patients treated surgically for OPLL as identified by the national Japanese Diagnosis Procedure Combination database, those patients undergoing posterior LAMP were significantly older and had significantly higher rates of comorbidities, including malignancy, cardiovascular disease, diabetes, renal failure, and cardiac failure. Additionally, some authors have posited that posterior approaches are generally associated with fewer complications and preserved ROM (e.g., LAMP), while also allowing the decompression of a larger number of segments in multilevel OPLL.<sup>18</sup> It is worth pointing out though that this may depend heavily on the surgeon and institution, other studies have found either equivocal or higher rates of complications associated with posterior approaches compared to anterior approaches.<sup>27,28,31</sup>

Two procedures constitute the bulk of posterior approaches offered in OPLL: (1) laminectomy and fusion (LF) and (2) LAMP. LAMP involves hinging open the lamina to increase canal diameter. It is characterized by less aggressive tissue disruption than that associated with LF, and can be performed using one of several different techniques.<sup>32</sup> The lack of instrumented fusion and laminectomy make the LAMP procedures better for preserving cervical ROM.<sup>33</sup>

Preserved cervical ROM, as well as the posterior approach in which OPLL is addressed indirectly,<sup>34,35</sup> do represent risks for OPLL progression, though the clinical importance of this remains to be fully elucidated. A recent meta-analysis of 11 studies directly comparing laminectomy and LAMP found that while radiographic OPLL progression was significantly higher in the LAMP group (62.5%; 95% CI, 55.3%–69.3%;  $I^2 = 45%$ ) compared to LF (7.6%; 95% CI, 2.4%–15.9%;  $I^2 = 0%$ ), the associated event rate of neurological decline was rare for both, and similar between the 2 groups: 8.3%, 95% CI, 3.7%–17.9%,  $I^2 = 60%$  vs. 2.8%; 95% CI, 1.3%–10.2%;  $I^2 = 0%$ .<sup>36</sup> This is in keeping with a large epidemiologic survey of over 3,000 patients with OPLL in Japan that revealed the vast majority of patients with OPLL required only one surgery, with a relatively small portion (11.1%) of patients undergoing repeat surgery.<sup>5</sup>

How choice of approach affects long-term neurological outcome is unclear in the literature,<sup>37,38</sup> and two recent meta-analyses comparing LAMP or LF and ADF offered differing conclusions, with one finding similar rates of JOA and recovery rates<sup>20</sup> and the other finding improved postoperative JOA scores and recovery rates with anterior approaches.<sup>38</sup>

## 3. Multilevel Approaches

Multilevel OPLL presents a significant treatment challenge.

One prospective nonrandomized study of 252 patients with multilevel cervical OPLL treated with either ACDF/ACCF or LAMP found overall complication rates as high as 41.33% in the anterior group and 50.98% in the LAMP cohort.<sup>27</sup> A recent meta-analysis comparing LF and LAMP procedures found the two to be equivocal in both clinical outcomes and surgical complications.<sup>39</sup> The clinical outcomes following traditional anterior and posterior approaches were adequate with patients demonstrating significant neurologic improvement with both approaches, but the greater risk for complications with these traditional approaches has sparked new considerations for patients with multilevel OPLL.

One such technique is the novel bridge crane “hoisting” technique, referred to as the “anterior controllable antidisplacement and fusion” (ACAF) procedure, first described by Lee et al.<sup>40</sup> This technique involves discectomy and anterior vertebral body resection of the involved levels, followed by placement of intervertebral grafts and an anterior plate to the vertebrae-OPLL complex (VOC) to form a “bridge.” Bilateral osteotomies are then performed at the widest portion of the multilevel OPLL mass to mobilize and isolate the VOC, which is then “hoisted” off the spinal cord by tightening of the cage screws. This technique allows for multilevel decompression to be achieved from an anterior approach without direct manipulation of the OPLL mass.<sup>41,42</sup> In comparison with a traditional ACCF approach, the ACAF technique demonstrated greater radiographic decompression, as well as significantly greater postoperative JOA scores and lower complication rates.<sup>41,43</sup> Another technique is the “skip” corpectomy and fusion (SCF) which aims to provide multilevel decompression while maintaining adequate stabilization.<sup>42</sup> This approach consists of C-4 and C-6 corpectomy, C-5 preservation, and C4-5 and C5-7 grafting with instrumentation of C-3, C-5, and C-7.<sup>42</sup> While this technique has not been analyzed extensively in patients with OPLL, a recent comparison of ACAF and SCF demonstrated better 6-month fusion rate and significantly fewer CSF leaks in patients treated with ACAF (0%) compared to SCF (16.7%,  $p < 0.05$ ).<sup>44</sup>

#### 4. Combined Approach

Generally, combined anterior and posterior approaches to the cervical spine are considered in situations of irreducible kyphotic deformity<sup>45</sup> or underlying metabolic derangements that can decrease the probability of successful fusion, including osteoporosis, diabetes or tobacco use.<sup>46</sup> In cases where 4 or more levels must be fused anteriorly, posterior instrumentation can be considered to augment successful arthrodesis.<sup>47</sup> In the OPLL

population, combined approaches should additionally be considered in patients with severe localized OPLL.<sup>32</sup> Arima et al.<sup>48</sup> reported success using a combined ACDF and partial OPLL mass resection with posterior cervical segmental decompression and fusion in a small case series of 5 patients with severe (>60% canal occupancy) localized OPLL. Patients with severe OPLL and extensive fixed kyphotic deformity that require long-segmental cervical fusion may warrant consideration for combined, 2-stage approaches. Lee et al.<sup>49</sup> recently described the use of a 2-stage 540° posterior and anterior-posterior (P-A-P) approach in a cohort of 18 patients with severe, multisegment OPLL and extensive kyphotic deformity. In this study, the P-A-P approach led to a significant improvement in both average C2-7 Cobb angle and postoperative JOA scores.<sup>49</sup>

#### 5. Surgical Approach Selection

One commonly used technique used to facilitate choice of the most appropriate approach to OPLL is the K-line, first described by Fujiyoshi et al.<sup>50</sup> in 2008. The K-line is an index incorporating both the kyphotic alignment of the cervical spine and thickness of the ossification and is defined as the line that connects the midpoints of the spinal canal at C2 and C7. K-Line (-) OPLL is defined as an OPLL foci whose peak exceeds the K-line and is associated with decreased C2-7 ROM and increased occupying ratio and extension/flexion ratio. In these patients, posterior surgical approaches have been shown to lead to inadequate posterior decompression of the spinal cord and significantly worse neurological outcomes.<sup>15</sup> This suggests that in K-line (-) patients, the anterior approach should be more highly considered, as shown by Koda et al.,<sup>51</sup> who achieved better JOA scores with anterior decompression and fusion than with posterior LAMP or decompression and fusion.

Alterations to the original K-line described by Fujiyoshi et al.<sup>51</sup> have served to improve clinical predictive power and usefulness. An additional study by Ijima et al.<sup>52</sup> showed that K-line measurements on radiograph versus CT images can vary significantly, and that for accuracy the K-line should be measured on a plain radiographs. A recent study by Lee et al.<sup>53</sup> introduced the kappa line, a modification of the K-line that is defined as a straight line connecting the midpoints of the spinal canal at one level above and one level below the decompressed segments. This new index had better predictive power with regards to neurologic recovery and cord compression following  $\leq 4$ -level LAMP than the K-line.

Ultimately, the choice of a surgical approach requires the consideration of numerous radiographic, technical, and patient-

**Table 3.** Algorithm for surgical approach selection adapted from Ha et al., 2016<sup>54</sup>

Approach	≤ 2 Levels Involved	≥ 3 Levels Involved
Anterior ADF, ACDF	Any occupying ratio	-
Laminoplasty	Only for occupying ratio >60%	If no local kyphosis is present
Combined anterior & posterior	-	If local kyphosis is present and occupying ratio is >60%
Posterior fusion	-	If local kyphosis is present and occupying ratio is ≤60%

Table summarizing the key portions of the algorithm described by Ha et al., 2016<sup>54</sup> that involves selecting surgical approach to based on the number of involved levels, the occupying ratio of the OPLL mass, and the presence of kyphotic deformity.

OPLL, ossification of the posterior longitudinal ligament; ADF, anterior decompression with floating; ACDF, anterior cervical decompression and fusion.

specific considerations and numerous algorithms exist to facilitate in this decision-making process and so are not reviewed extensively herein. One such algorithm, proposed by Ha et al.<sup>54</sup> (adapted in Table 3), takes into account the extent of OPLL disease (number of levels involved) and occupying ratio to aid in approach selection.

## 6. Complications

The incidence of complications associated with the operative treatment of OPLL may depend on a variety of factors including surgical approach, surgeon experience, and the presence of general systemic as well as neurologic comorbidity, including dural ossification (DO). The reported incidence of complications varies widely, ranging from 5.2% to 57.6%, with a cumulative rate of 21.8% according to one review of the literature, and 21.48% according to the multicenter AOSpine International Study of patients treated surgically for DCM.<sup>21,31</sup> Multivariate analysis of the AOSpine International Study found that OPLL was in fact a significant risk factor for perioperative complications in patients undergoing surgical treatment of DCM (OR, 1.75;  $p = 0.040$ ).<sup>55</sup>

### 1) Dural tear & CSF leak

Dural tear (DT) is a known adverse event associated with anterior approaches to OPLL. In extreme cases, these may lead to need for reoperation, pseudomeningocele formation, persistent fistula formation, or meningitis, and may even require permanent CSF diversion to address definitively.<sup>56</sup> The incidence of DT in cervical spine surgeries has repeatedly been reported to be elevated when OPLL is present. Analysis of a nationwide database of all incidental DTs in cervical spine surgery in the United States in 2009 revealed a significant association between the presence of OPLL and DT (OR, 58.36; 95% CI, 14.75–230.82;  $p < 0.001$ ).<sup>57</sup> Similarly, a 2017 meta-analysis of all DCM surgeries revealed a higher rate of CSF leak with OPLL surgeries (12.2%;

95% CI, 6.3%–17.8%), and the AOSpine International Study found a higher frequency of DTs in patients with OPLL (5.2%) compared to other DCM pathologies (2.0%) though this difference was not significant ( $p = 0.076$ ).<sup>21,58</sup> The increased risk of DT in OPLL is attributable to the intimate adhesion of the dura to the ossified PLL mass. This includes the presence of DO, in which the ossification of the PLL extends into the dura itself. DO is estimated to affect between 15.3%–29.0% of patients with OPLL, and is most frequently identified on axial CT via the “double-layer” sign, which has a sensitivity of 55% and a specificity of 96.9%.<sup>59,60</sup> In a small, single institution study of 126 patients, Yu et al.<sup>61</sup> demonstrated DT in 64.6% of patients with evidence of DO on axial CT.

According to a 2016 meta-analysis of OPLL surgeries, the incidence of DT was estimated to be as high as 31% for anterior approaches, considerably higher than that of posterior approaches (9.3%; OR, 1.90; 95% CI, 1.08–3.36;  $p < 0.05$ ).<sup>37</sup> A recent analysis of 1,192 propensity-score matched pairs of patients found the rate of CSF leak to be significantly higher in ADF (2.4%) procedures compared to posterior LAMP (0.4%,  $p < 0.001$ ).<sup>30</sup> However the results are not entirely uniform, with some studies failing to demonstrate higher rates of DTs and CSF leaks during anterior approaches. For example, a nationwide analysis of DTs by Yoshihara and Yoneoka<sup>57</sup> noted posterior-only approaches (OR, 2.59; 95% CI, 1.70–3.96;  $p < 0.001$ ) and combined approaches (OR, 3.36; 95% CI, 1.99–5.68;  $p < 0.001$ ) to carry the highest risk for DT. In a nonrandomized prospective study of 150 patients at a single institution, Hou et al.<sup>27</sup> demonstrated similar rates of CSF leak between ADF and LAMP in multilevel OPLL (3.33% vs. 2.94% respectively,  $p > 0.05$ ).

Considering the association of OPLL with DTs, and evidence that suggests there exists a higher rate of failed DT treatment in patients with OPLL, a number of strategies have been devised to minimize the risk of DT in the treatment of OPLL.<sup>47</sup> The first of these, aptly referred to as the “anterior floating method,” in-

volves meticulous thinning of the OPLL with a high-speed drill, often under microscopic visualization, followed by release of the ossified mass from the vertebral bodies, without complete resection of the tissue from the dura.<sup>24</sup> One retrospective study of 144 patients treated with the ADF method demonstrated a CSF leak rate of (6.3%) which is comparatively lower than previous reports of incidence of CSF leak.<sup>62</sup> Nonetheless, it should be noted that a recent meta-analysis of studies comparing ADF and posterior LAMP found higher rates of CSF leak in the ADF population (15.74%) compared to LAMP (5.21%), though these differences were not significant. To further minimize the risk of complications with ADF, Yoshii et al.<sup>63</sup> have proposed use of intraoperative CT along with the ADF procedure. In a small RCT of 25 patients undergoing ADF using intraoperative CT, the enhanced imaging modality was found to confer better postoperative outcome and a lower rate of complications compared to ADF without intraoperative CT. More recently, the ACAF technique was used to treat to a small cohort of 28 patients with OPLL and evidence of DO, and successfully demonstrated a significantly lower rate of CSF leak with ACAF (3.6%) compared to anterior corpectomy (22.6%,  $p < 0.01$ ).<sup>43</sup>

When there is the absence of dura due to its ossification and incorporation with the PLL, removal may result in a CSF leak. Once the leak is identified intraoperatively, various techniques can be used to repair the defect and prevent further complications. Primary watertight repair remains ideal, but this is not always practical from the anterior cervical approach. Various other on-lay strategies, including the use of fibrin glue and graft materials, can be employed when primary repair is not attainable.<sup>64-67</sup> An extensive review of the literature by Mazur et al.<sup>66</sup> suggested that the use of CSF diversion via lumbar drain or shunt in dural leaks treatment following OPLL surgery has a high success rate, ranging from 83%–100%. The use of other more invasive forms of CSF diversion has only been reported in one study that found 5 of 82 patients treated with anterior corpectomy for multilevel OPLL required wound-peritoneal or lumboperitoneal shunting.<sup>68</sup> While CSF diversion and upright bedrest may constitute the dogma of dural leak management, Moon et al.<sup>69</sup> recently reported on a small case series of 7 patients with OPLL and dural leaks that were managed without lumbar drainage or bedrest. Of the 4 patients who developed pseudomeningocele, early ambulation led to spontaneous resorption in 2 patients, and early stabilization in the others. While more data is needed, this finding challenges the traditional notion of management of dural leaks.<sup>64</sup>

## 2) C5 palsy

The etiology of the so-called postoperative “C5 palsy” has yet to be definitively established, however it is a phenomenon that is widely acknowledged after cervical spine surgery and is certainly not unique to surgeries for OPLL. Onset of C5 palsy is typically within the first week of surgery, but may appear as early as immediately postoperatively, or as late as 4 weeks postoperatively.<sup>70</sup> Two potential explanations of the pathophysiology of C5 palsy exist including a tethering phenomenon, in which posterior decompression leads to spinal cord shift with resultant traction on the spinal cord and/or nerve roots, and alterations in spinal cord perfusion in the context of impaired autoregulation mechanisms caused acutely at the time of decompression.<sup>71</sup>

Among approaches for the OPLL population, posterior approaches have consistently reported higher rates of C5 palsy compared to anterior approaches.<sup>31,37</sup> The extent to which C5 palsy following OPLL surgery can be attributed to the surgical procedure itself, or the presence of OPLL, is not clear. While a recent meta-analysis of 61 studies of C5 palsy in all forms of DCM surgeries suggests that incidence of C5 palsy is higher in patients with OPLL (8.1%) compared to other DCM patients (4.8%), the AOSpine study found equivocal rates of C5 palsy between surgeries for OPLL compared to surgery for other forms of DCM (1.48% vs. 0.58%,  $p = 0.32$ ).<sup>21,71</sup> According to Wang et al.,<sup>71</sup> the incidence of C5 palsy is higher in patients with OPLL undergoing LAMP (5.5%) or anterior cervical decompression and fusion (8.1%) compared to patients with other forms of DCM (4.7%, 3.1%, respectively). Furthermore, Singhatanadgige et al.<sup>33</sup> found that when only considering patients with OPLL, the incidence of C5 palsy was higher following LF (9.6%–25%) compared to LAMP (0%–8%).

As was true for DTs related to OPLL surgery, strategies have been introduced aiming to reduce the incidence of C5 palsy, although much of this is without objective evidence in favor of improved outcome. Methods have been sought to better predict the onset of C5 palsy postoperatively. Takeuchi et al.,<sup>72</sup> for example, recently proposed that larger C5 nerve root cross-sectional area on preoperative cervical ultrasonography was capable of successfully predicting postoperative C5 nerve root palsy.

## 3) Axial pain

Axial neck pain may be the most frequently reported complication of OPLL surgery, with reports of its incidence ranging anywhere from 16%–48%.<sup>27,31,38</sup> Despite this, reliable data on axial pain is significantly lacking, with only a few studies reporting on its incidence quantitatively. Most published reports

indicate that anterior approaches are less likely to be associated with significant axial pain, regardless of the underlying diagnosis.<sup>27,38</sup> For example, Wang et al.<sup>58</sup> found the incidence of axial pain among 23 studies of decompression for DCM, including OPLL, to be 15.6% (95% CI, 11.7%–19.5%), with the highest incidence being in LAMP (22.2%; 95% CI, 14.1%–29.3%) and LF (23.2%; 95% CI, 15.8%–31.3%). Evidence to the contrary includes the AOSpine study, which found a significantly higher proportion of OPLL patients reporting new onset neck pain following surgery compared to patients with other forms of DCM.<sup>21</sup>

#### 4) *Dysphagia, dysarthria, and hoarseness*

Dysphagia, dysarthria, and hoarseness are well-known complications of anterior cervical procedures, and are related to manipulation and retraction of the esophagus, or direct injury to the recurrent laryngeal nerve. A recent analysis of 1,192 propensity-score matched pairs of patients with OPLL found the rate of dysphagia to be higher in ADF (2.4%) compared to LAMP procedures (0.4%,  $p < 0.001$ ).<sup>30</sup> Wang et al.<sup>58</sup> reported an incidence of dysphagia of 1.4%–58.1% with an average of 16.8% (95% CI, 13.6%–19.9%) across 38 studies of DCM. Similarly, the incidence of hoarseness ranged from 0.6%–60.9% with an average incidence of 4.0% (95% CI, 2.3%–5.7%). The incidence of both dysphagia and hoarseness were reported to be higher with ACDF and ACCF procedures, but did not appear to be related to the presence of OPLL.<sup>58</sup> The multicenter AOSpine study found similar results, with the rate of dysphagia being not significantly affected by the presence of OPLL.<sup>21</sup> Subgroup analysis of patients with all types of DCM who experienced postoperative dysphagia in the AOSpine International and North America studies identified a number of risk factors for postoperative dysphagia, including the presence of endocrine comorbidities, and greater number of decompressed segments.<sup>73</sup>

#### 5) *Hematoma*

Hematoma formation, both surgical site hematomas, and remote intracranial hematomas, are well-described complications occurring after spinal surgery. Both are potentially life-threatening complications that often require emergent action.<sup>31,74</sup> Estimates of the prevalence of postoperative surgical site epidural hematoma in cervical spinal surgery for DCM range from 0.5%–5.3% with an overall incidence of 1.1% (95% CI, 0.7%–1.5%).<sup>58</sup> The nonrandomized prospective study by Hou et al.<sup>27</sup> found the overall rate of epidural hematoma formation in multilevel OPLL surgery to be similar, at 1.98%, with no significant differ-

ence in the rate with anterior surgery (2.0%) compared to posterior surgery (1.02%,  $p > 0.05$ ). The meta-analysis conducted by Feng et al.<sup>37</sup> found relatively higher rates of hematoma formation in both anterior (5.1%) and posterior (4.7%) approaches compared to other studies, though the difference between the 2 groups remained statistically insignificant. In the cohort of patients undergoing the “bridge crane hoisting” ACAF as described by Yang et al.,<sup>75</sup> no epidural hematoma formation was reported. While limited data is available on the development of retropharyngeal hematoma, one single institution study of 2,375 cervical spine surgery patients found that the presence of OPLL conferred an increased risk for retropharyngeal hematoma formation (relative risk [RR], 6.8; 95% CI, 2.3–20.6).<sup>74</sup>

#### 6) *Hardware complications*

Complications related to use of hardware, include graft subsidence, screw migration, pseudarthrosis, among others, do not appear to have a significantly higher incidence in patients with OPLL compared to other forms of DCM.<sup>21,58</sup> It should be noted that while some studies have found higher rates of pseudarthrosis and hardware-related complications in patients treated with anterior approaches,<sup>31,37,76</sup> more recent reports on comparisons of anterior and posterior approaches have failed to replicate these findings.<sup>21,27</sup>

#### 7) *Other neurologic complications*

The reporting of neurologic complications other than C5 palsy is limited in the literature. A meta-analysis of studies on OPLL found the incidence of postoperative neurologic deficit to be 8.4% (1,558 patients) though C5 palsy accounted for half of reported deficits (4.2%).<sup>31</sup> Most of these patients spontaneously recovered function in the postoperative period. Another meta-analysis comparing anterior and posterior approaches found 9.3% of patients treated with the posterior approach had postoperative radiculopathy compared to no patients treated with the anterior approach.<sup>37</sup> Employing intraoperative neuromonitoring (IONM) techniques may reduce the incidence of neurologic complications associated with these procedures. Changes in somatosensory evoked potential (SEP) can signal impending spinal cord injuries, and Epstein et al.<sup>69</sup> showed a reduction in neurologic deficits associated with SEP monitoring. Additionally, a recent multicenter analysis by Yoshida et al.<sup>77</sup> found a rescue rate of 82.1% when IONM was used during surgical correction of cervical OPLL.



### 8) Other surgical complications

The limited evidence comparing the incidence of other miscellaneous surgical complications, such as deep vein thrombosis, pulmonary embolism, pneumonia, and urinary tract infection do not indicate an association with OPLL.<sup>30,55</sup> Interestingly, the AOSpine International Study did find a higher rate of superficial surgical site infections in patients with OPLL (6.67%) compared to other forms of DCM (1.16%,  $p = 0.002$ ).<sup>55</sup> Additionally, propensity-matched scoring revealed a significantly higher rate of surgical site infection in patients treated with LAMP (3.4%) compared to ADF (2.0%,  $p = 0.033$ ).<sup>30</sup> However, a meta-analysis of cervical surgeries found the reported rate of infection to range widely from 0.4% to 54.6% with no observable relation to the presence of OPLL.<sup>58</sup>

## CONCLUSIONS

OPLL is an uncommon condition but potentially devastating disease if left untreated. Surgical decompression is indicated for moderate to severe, or progressive symptoms. This can be achieved via a variety of different approaches.<sup>20</sup> Posterior approaches, including LF and LAMP, can be used to treat a wide range of OPLL disease severities making it the most popular approach to surgical OPLL correction worldwide.<sup>21</sup> Anterior approaches to OPLL are more technically demanding, but provides for direct decompression of the OPLL mass, and may be associated with better postoperative outcomes and less disease progression.<sup>28</sup> Generally, surgical correction of OPLL is associated with higher rates of complications, and its presence is considered a significant risk factor for perioperative complications in DCM surgeries.<sup>55,58</sup> In particular, the rates of intraoperative dural leaks and postoperative C5 palsy have both been reported to be higher in patients with OPLL compared to other DCM pathologies.<sup>21,71</sup> These complications may be specific to certain approaches, however, with rates of DT being higher with anterior approaches and C5 palsy being higher with LF posterior approaches.<sup>30,31,33,37</sup> Several techniques specific to OPLL surgeries have been developed to combat the incidence of these complications including the ADF and the ACAF procedures.<sup>24,41</sup> Aside from some literature substantiating a higher rate of wound infection in the OPLL population, the incidence of other complications, including dysphagia, hardware failure, other neurologic deficit, venous thromboembolism, pneumonia, and urinary tract infection do not appear to be altered by the presence of OPLL.

## FUTURE DIRECTIONS

This article represents a review of the literature on the surgical correction of OPLL and its associated complications. We find that the low incidence of OPLL makes for a paucity of level-I evidence comparing surgical approaches to OPLL, creating a dire need for randomized-controlled trials comparing anterior and posterior approaches to surgical correction of OPLL.

## CONFLICT OF INTEREST

Dr. Christopher Maulucci serves as a consultant for Globus Medical. The other authors have nothing to disclose. The views expressed in this document are those of the authors and do not necessarily reflect the official policy or position of the Department of the Army, the Department of Defense, nor the U.S. Government.

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