OSSIFICATION of the posterior longitudinal ligament (OPLL) is most commonly found in men, the elderly, and Asian patients. There are many diseases associated with OPLL, such as diffuse idiopathic skeletal hyperostosis, ankylosing spondylitis, and other spondyloarthropathies. Several factors have been reported to be associated with OPLL formation and progression, including genetic, hormonal, environmental, and lifestyle factors. However, the pathogenesis of OPLL is still unclear. Most symptomatic patients with OPLL present with neurological deficits such as myelopathy, radiculopathy, and/or bowel and bladder symptoms. There are some reports of asymptomatic OPLL. Both static and dynamic factors are related to the development of myelopathy. Plain radiography, CT, and MR imaging are used to evaluate OPLL extension and the area of spinal cord compression. Management of OPLL continues to be controversial. Each surgical technique has some advantages and disadvantages, and the choice of operation should be made case by case, depending on the patient’s condition, level of pathology, type of OPLL, and the surgeon’s experience. In this paper, the authors attempt to review the incidence, pathology, pathogenesis, natural history, clinical presentation, classification, radiological evaluation, and management of OPLL. (DOI: 10.3171/2010.11.FOCUS10276)

Key Words: ossification, posterior longitudinal ligament, spine

Abbreviations used in this paper: ACDF = anterior cervical disectomy and fusion; BMD = bone mineral density; BMP = bone morphogenetic protein; CSM = cervical spondylotic myelopathy; DISH = diffuse idiopathic skeletal hyperostosis; JOA = Japanese Orthopedic Association; OALL = ossification of the anterior longitudinal ligament; OLF = ossification of ligamentum flavum; OPLL = ossification of the posterior longitudinal ligament; TGF = transforming growth factor.

Methods

The PubMed databases were searched for publications from January 2000 through August 2010 using the MeSH terms “OPLL” and “ossification of posterior longitudinal ligament.” The search was limited to articles in the English language. Related reference sections of recent articles were reviewed and pertinent articles identified. Full-texts manuscripts of all articles were obtained and reviewed. Radiographic images from the senior author’s institution are also included.

Results

Incidence

The incidence of OPLL was reported by Tsuyama et al. The incidence is 2.4% in Asian populations and 0.16% in non-Asian populations, with the highest rates in Japan. OPLL is twice as common in men as in women, and symptomatic OPLL usually presents in the 5th to 6th decade of life. Most studies of OPLL are reported from Asian countries, but anecdotal reports of OPLL cases in European countries also exist in the literature. Maiuri et al. reported on 8 Italian patients with cervical spine stenosis due to OPLL.
OPLL has been reported to be associated with other musculoskeletal diseases such as DISH (or Forestier disease),36,26,27,30,51,60,69,81,109 ankylosing spondylitis, and other spondyloarthropathies.54,108 Resnick et al.109 found OPLL in 50% of patients with DISH. In 2009, Kawabori et al.34 reported a rare case of DISH with continuous-type OPLL at C2–4 that presented with cervical myelopathy from C-1 posterior tubercle impingement. Multilevel fusion of the subaxial cervical spine from OPLL caused hypermobility at C1–2 and may lead to ligamentous damage and subsequent C-1 posterior tubercle impingement. In Japanese cases reported by Tsuyama,126 2% also had ankylosing spondylitis. Tyrrell et al.128 reported a case of OPLL in a patient with Down syndrome. A high incidence of OPLL (20%) has been reported by Matsunaga et al.75 in patients with schizophrenia. The authors also reported OPLL in dizygotic twins with schizophrenia.

Pathology

OPLL is believed to form through endochondral ossification. McAfee et al.81 described the histopathology of OPLL, which is composed largely of lamellar bone with mature Haversian canals. Ultrastructural study of the ligament flavum in patients with OPLL revealed atrophic elastic bundles with a 2-layer structure, disappearance of microfibrils, irregular alignment of collagen fibrils, and many extracellular plasma membrane-invested particles that resemble matrix vesicles.90

Pathogenesis

The pathogenesis of OPLL remains poorly understood. There is some evidence that ligament cells from patients with OPLL have osteoblast-like characteristics. Ishida and Kawai41 studied cell lines from nonossified sites in patients with OPLL and found that they have high alkaline phosphatase activity, response to calcitonin, and calcitriol. Parathyroid hormone and dinoprostone can also stimulate an increase in cyclic adenosine monophosphate in these cell lines. There are many proposed genetic, hormonal, environmental, and lifestyle factors that relate to pathogenesis and progression of OPLL, but most of these theories are still controversial.

An immunohistochemical study of extracellular matrix components in the twy (tiptoe walking Yoshimura) mouse, an animal model for the study of OPLL, shows that degeneration and subsequent herniation of the nucleus pulposus is the potent regional factor that initiates OPLL formation. At 14 weeks, the discs herniated into the thickened posterior longitudinal ligament, then cartilaginous tissue appeared in the posterior longitudinal ligament as if to repair the intervertebral disc degeneration.35

Hypertrophy of the posterior longitudinal ligament is believed to be an early stage of OPLL. Histological and biochemical study of hypertrophy of the posterior longitudinal ligament shows hyalinoid degeneration, proliferation of chondrocytes and fibroblast-like spindle cells, infiltration of vessels and small ossification, and staining by BMP, TGF-β, and proliferating cell nuclear antigen, which are all similar to OPLL.112

Genetic Factors. Patients with OPLL are most commonly found in Asian populations, so genetic factors are considered to be a factor in OPLL development. Tanabe et al.119 reported a case of OPLL in the thoracic spine; this patient had a brother with the same disease, also in the thoracic spine. Genetic factors are believed to contribute to OPLL development. Many collagen genes have been studied, including human collagen α2 gene (COL11A2). Koga et al.61 showed that this gene, located on chromosome 6p close to the human leukocyte antigen region, is strongly associated with OPLL. Retaining exon 7 together with removal of exon 6 observed in intron 6-(4A) in the COL11A2 gene could play a protective role in the ectopic ossification process.70 Maeda et al.71 reported a sex-specific association of the COL11A2 haplotype with OPLL in male patients. However, a recent study by Horikoshi et al.78 could not reproduce the association between this gene and OPLL.

A single nucleotide polymorphism in intron 32(−29) in the collagen 6A1 gene (COL6A1) on chromosome 21q22.3 is associated with OPLL.120,123 Kong et al.144 studied the Han Chinese population and also found a significant association of COL6A1 with OPLL. They demonstrated that 3 single nucleotide polymorphisms, including promoter (-572T), intron 32(−29), and intron 33(+20), are significantly associated with OPLL and OLF.

Okawa et al.101,102 identified a mutation of the NPPS gene as the cause of tiptoe walking condition in the twy mouse. Nucleotide pyrophosphatase is a membrane-bound glycoprotein believed to produce inorganic pyrophosphate, a major inhibitor of calcification and mineralization. Some evidence suggests that NPPS gene mutation is associated with OPLL development.65,91 In a later study by Tahara et al.,34 the authors showed that NPPS and leptin receptor genes do not promote an increased susceptibility to OPLL, but are associated with the extent of heterotopic ossification. Horikoshi et al.38 also could not demonstrate the association between the NPPS gene and OPLL.

Human retinoic X receptor β, TGF3, BMP4, FF variant of vitamin D receptor gene,58 promyelocytic leukemia zinc finger gene, and Runt-related transcription factor 2 (RUNX2) are linked to OPLL with anecdotal evidence. Angiopoietin-1, a downstream of RUNX2, may play an important role in ectopic calcification.57

Hormonal Factors. Bone morphogenetic protein, a substance with the ability to induce ectopic bone and cartilage formation, is believed to play an important role in the pathogenesis of OPLL. Bone morphogenetic protein receptors increased in ossified ligament tissue in patients with OPLL.41 Bone morphogenetic protein-2 stimulates differentiation of ligament cells in patients with OPLL and induces ossification by increasing alkaline phosphatase activity and stimulating DNA and procollagen Type I carboxyl-terminal peptide synthesis.65 The TC and CC genotypes in exon 3(−726) T/C in the BMP-2 gene of male Han Chinese patients have a genetic susceptibility to OPLL in the cervical spine.130 Wang et al.129 demonstrated an association between the Ser37Ala (T/G) polymorphism and the occurrence of OPLL. They also showed significant linkage between the Ser87Ser (A/G) polymorphism and the extent of cervical ossification.
Ossification of the posterior longitudinal ligament

Transforming growth factor-\(\beta\) has been studied in the literature. The T869→C polymorphism of the TGF-\(\beta\)I gene is a genetic determinant of a predisposition to OPLL.\(^{48}\) In a later study, Kawaguchi et al.\(^{52}\) demonstrated that the TGF-\(\beta\)I polymorphism is not associated with OPLL development, but rather a factor related to the extent of ossification. Patients with the C allele frequently have OPLL in the cervical, thoracic, and/or lumbar spine.

In a study of serum biomarkers for OPLL, Eun et al.\(^{20}\) showed that 8 biomarkers were upregulated in the sera of OPLL patients: 1) PRO2675, 2) human serum albumin in a complex with myristic acid and triiodobenzoic acid, 3) an unknown protein, 4) chain B of the crystal structure of deoxy-human hemoglobin \(\beta6,5\) proapolipoprotein, 6) albumin protein, 7) retinol binding protein, and 8) chain A of human serum albumin mutant R218h complexed with thyroxine, whereas \(\alpha1\)-microglobulin/bikunin precursor was downregulated. Matsui et al.\(^{73}\) demonstrated increased serum procollagen Type I carboxyl-terminal peptide and intact osteocalcin in patients with OPLL. These markers also increased in concert with the progression of OPLL without statistical significance. Cerebrospinal fluid analysis in patients with OPLL and CSM showed high levels of interleukin-8.\(^{42}\)

Non–insulin-dependent diabetes mellitus has been suggested as a risk factor of OPLL.\(^{59}\) Li et al.\(^{67}\) showed increased expression of insulin receptors, proliferation of rat spinal ligament cells, and induction of osteogenic differentiation through the PI3-K/Akt pathway induced by insulin. Insulin-like growth factor-I induces histological change and elevation of alkaline phosphatase activity in OPLL cell lines much more than in non-OPLL cells.\(^{75}\)

OPLL is a disease that results in increased bone formation in ligament tissue, and there is some evidence showing correlation between OPLL and increased overall BMD. In several studies, patients with OPLL had higher BMD than the non-OPLL controls,\(^{34,53,134}\) but BMD may decrease in patients with advancing OPLL.\(^{87}\) Aita et al.\(^{2}\) studied histomorphometry of the iliac bone in patients with OPLL and found no significant differences between OPLL and control groups. They speculated that stage of OPLL and disuse atrophy may be the responsible factors.

High serum levels of menatetrenone in male patients\(^{55}\) and activin in male and female patients\(^{54}\) have been investigated and correlated with OPLL formation. Tumor necrosis factor \(\alpha\)-stimulated gene-6 suppresses osteoblastic differentiation induced by BMP-2 and osteogenic differentiation medium.\(^{124}\) The author of this study suggested that this is a plausible target for therapeutic intervention in OPLL.

**Environmental Factors.** Mechanical stress in ligaments of the spine has been investigated as a cause of OPLL development and progression.\(^{22}\) Prostacyclin synthase levels in ligament cells from OPLL patients have been shown to be elevated after applying mechanical stress and induced osteogenic differentiation via the PGI2/cyclic adenosine monophosphate pathway.\(^{97}\) Mechanical stress also induces mRNA expression of alkaline phosphatase, osteopontin, BMP-2, BMP-4, BMP receptors,\(^{121}\) and mRNA expression of Cbfa1, Type I collagen, osteocalcin, integrin \(\beta1,\) and endothelin-1.\(^{47}\) The P2Y1 purinoceptor subtypes, intensively expressed in OPLL cells, responded to mechanical stress–induced extracellular adenose triphosphate, which stimulated OPLL progression.\(^{110}\)

Frequent consumption of pickles, nondaily consumers of rice,\(^{100}\) family history of myocardial infarction, high body mass index at age 40, long working hours, and working night shifts\(^{58}\) were associated with increased risk of OPLL. On the other hand, frequent consumption of chicken and soy products\(^{103}\) and good sleeping habits (6–8 hours/night) in the prime of life may decrease the risk of OPLL.\(^{531}\)

**Natural History**

Symptomatic OPLL is usually detected in elderly patients. There have been several studies that investigated the natural history of OPLL. Chiba et al.\(^{10}\) described computer-assisted measurement of the size of OPLL. They reported excellent inter- and intraobserver reliability of this method with 98% accuracy for detecting OPLL progression. Thereafter, they applied this method to 131 patients with cervical OPLL who underwent posterior decompression at 13 institutions. The rate of OPLL progression was 56.5% at 2 years and was more common in younger patients with continuous- and mixed-type OPLL.\(^{12}\) Murakami et al.\(^{69}\) reported a case of cervical OPLL in a 67-year-old man with more than 26 years follow-up. They found that the rate of OPLL progression varied during this period. The rate of progression was 2.2, 8.8, and 2.0 mm/year from 1–4, 4–8, and 8–10 years after the first visit, respectively. After 10 years, there was no evidence of OPLL progression.

Hori et al.\(^{56,57}\) investigated the progression of OPLL in both longitudinal axis and thickness in 55 patients with at least a 5-year follow-up period. They found that progression was marked in younger patients with continuous- or mixed-type OPLL, consistent with the results of Chiba et al.\(^{15}\) According to progression on the longitudinal axis, the patients with continuous- or mixed-type OPLL were classified according to age. The patients 40–49 years of age showed peak progression at greater than 1 year, whereas the patients older than 50 showed peak progression during the first year of follow-up. The authors suggested that OPLL might show a rapid progression in the 4th decade of life and that the progression gradually decreases in the 5th or 6th decade. For progression in thickness, the other factor that influences the progression is C-3 involvement; the progression of OPLL was frequently observed at levels C2–4.

Long-term follow-up of 450 patients with OPLL was reported by Matsunaga et al.\(^{77}\) in 2004. All patients were followed-up for at least 10 years, with a mean follow-up period of 17.6 years. Only 17% of patients without myelopathy at the first visit developed myelopathy during the follow-up period. The myelopathy-free rate in these patients was 71% after 30 years according to Kaplan-Meier analysis. The researchers suggested that prophylactic surgery in patients without symptoms of myelopathy is unnecessary. This same group of authors\(^{76}\) studied predictors for development of myelopathy in 156 patients with...
OPLL from 16 spine institutes with an average follow-up period of 10.3 years. They found that both static and dynamic factors were related to the development of myelopathy. All 39 patients with more than 60% spinal canal stenosis on plain radiography developed myelopathy. Range of motion was significantly greater in patients with myelopathy. Of 15 patients with trauma-induced myelopathy, 13 had mixed-type and 2 had segmental-type OPLL.

Clinical Presentation

Clinical presentation of OPLL depends on the size of the OPLL, spinal canal diameter, and range of motion of the spine. Some patients have no symptoms, but others present with neurological deficits such as radiculopathy, myelopathy, and in severe cases, bowel and bladder symptoms. The onset of symptoms is usually gradual, but there are also some reports of patients with trauma-induced sudden onset myelopathy.

Classification

The Investigation Committee on OPLL of the Japanese Ministry of Public Health and Welfare described the OPLL classification that is most widely used in the literature.126 Based on lateral plain radiography, cervical OPLL can be classified into 4 types (Fig. 1): continuous, segmental, mixed, or circumscribed type. Continuous type is classified as a long lesion extending over several vertebral bodies. Segmental type is classified as one or several separate lesions behind the 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.

Radiological Evaluation

Plain radiography is the simplest method for detecting OPLL but it has some limitations. Chang et al.6 reported low inter- and intraobserver reliability of lateral radiography as a tool for OPLL classification, particularly for continuous-type OPLL. The inter- and intraobserver kappa values were only 0.51 and 0.67, respectively. They emphasized the importance of 2D or 3D reconstructed images to overcome this problem.

Computed tomography and/or myelography are useful tools for detecting and accurately locating OPLL. The exact dimensions and extent of cervical canal stenosis are precisely depicted on CT. Figure 2 shows CT scans of patients with OPLL. A mushroom or hill shape on an axial CT scan typifies OPLL, and a sharp radiolucent line between the posterior vertebral body and ossified ligament is a also characteristic feature.113

Anterior decompression of OPLL in patients with associated dural ossification is more harmful compared with those without dural ossification because the incidence of new neurological deficits and CSF leakage is higher. A CT scan can be useful for detection of dural ossification. Mizuno et al.86 retrospectively reviewed the relationship between dural ossification and preoperative imaging. They found that bone window CT scans were the most useful method for detecting dural ossification, whereas MR imaging was ineffective in recognizing dural ossification. Of the 4 cervical OPLL types, the non-segmental type was most likely to be associated with dural ossification. Hida et al.31 classified bone window CT images to detect dural defect into 2 types: double- and single-layer sign. Single-layer sign was defined as a large focal mass of uniformly hyperdense OPLL. Double-layer sign was defined as anterior and posterior rims of hyperdense ossification separated by a central hypodense mass (the hypertrophied but nonossified posterior longitudinal ligament). Dural defects during surgery were detected in 10 of 12 patients with double-layer sign compared with only 1 of 9 patients with single-layer sign. Epstein19 applied this classification to her patients and demonstrated a dural defect in 1 of 12 patients with single-layer sign and an irregular C angular configuration compared with 1 of 4 patients with double-layer sign. She concluded that double-layer sign is more pathognomonic than single-layer sign for dural penetration. Smooth-layer sign indicated a clean dural plane with a low incidence of dural defect.

Min et al.84 studied 197 patients with cervical OPLL.
Ossification of the posterior longitudinal ligament

who underwent anterior decompression and fusion. There were signs of dural penetration in 30.5% of patients. These signs were more common in nonsegmental OPLL. Dural defects were detected in 20 (52.6%) of 38 patients with double-layer sign compared with 3 (13.6%) of 22 patients with single-layer sign. They also demonstrated a positive correlation between thickness of the central hypodense mass and the possibility of a dural defect. Signs of dural ossification are even more common in thoracic OPLL. Min et al. reported an 80% dural ossification rate in patients with thoracic OPLL. Dural defects were detected in 6 of 10 patients with double-layer sign and 3 of 6 patients with single-layer sign. Although most signs of dural ossification were detected in nonsegmental OPLL in the cervical spine, they were detected in both segmental and nonsegmental OPLL in the thoracic spine. The

Fig. 2. Computed tomography scans showing OPLL in different locations. A and B: Sagittal reconstructed images show large OPLL at C2–5 (arrow). C–E: Axial images of the cervical spine show a large OPLL occupying more than 50% of the spinal canal (arrows). F: Sagittal reconstructed image of the thoracic spine shows a mixed-type OPLL. The spinal canal is narrowest at T2–3 (arrow). G: Axial CT image shows large a OPLL occupying more than 80% of the spinal canal (arrow).
studies that correlated CT images and dural defect are summarized in Table 1.

Magnetic resonance imaging is inadequate for diagnosing small ossified lesions in the spinal canal, but is a sufficiently sensitive tool for detecting soft tissue abnormalities. A characteristic OPLL, signal hypointensity on both T1- and T2-weighted MR imaging, is shown in Fig. 3. In a study by Koyanagi et al., associated disc protrusion was found at maximum compression level in 60% of patients with cervical OPLL. Its presence is more common in segmental OPLL, with an incidence of 81%. The authors concluded that MR imaging is helpful for determining the actual level of spinal cord compression and for suggesting the optimal method of surgical treatment. Signal hyperintensity T2-weighted changes of the spinal cord are correlated with more severe neurological deficit. Yagi et al. demonstrated a positive correlation between postoperative expansion of the high signal intensity area of the spinal cord and poor neurological outcomes of patients with cervical OPLL. A risk factor for the expansion of the high signal intensity area was spinal instability.

The cross-sectional shape of the spinal cord at the level of maximum compression was classified as boomerang, teardrop, or triangular by Matsuyama et al. These investigators found that the recovery rate of patients with the triangular shape was worst, whereas the teardrop shape was best, and the boomerang shape was intermediate. After surgical intervention, triangular shape spinal cords showed the least expansion, which correlated with poor outcome.

Concurrent OPLL at multiple locations has been described. In a study of 68 patients with cervical OPLL by Park et al., thoracic tandem ossification was found in 23 cases (33.8%); 21 had thoracic OLF, 5 had thoracic OPLL, and 3 had both combined. The authors suggested performing simultaneous thoracic spine studies in patients undergoing cervical OPLL surgery.

Management

The mainstay treatment of OPLL is surgical decompression. Although there is a lot of research about OPLL formation and progression, such as genetic studies, growth factors, cytokines, and environmental factors, effective medical treatment for OPLL is still lacking. Most are only symptomatic treatments such as pain medication, topical agents, antiinflammatory drugs, antidepressants, anticonvulsants, nonsteroidal antiinflammatory drugs, and opioids.

Surgical Management of Cervical OPLL. The most common location of OPLL is at the cervical spine. There are several reports of surgical management of cervical OPLL with options including the posterior approach (laminectomy, laminectomy with fusion, laminoplasty, and open-door and double-door laminoplasty), the ante-
Ossification of the posterior longitudinal ligament

TABLE 2: Summary of advantages and disadvantages of surgical procedures for cervical OPLL*

<table>
<thead>
<tr>
<th>Surgical Procedure</th>
<th>Advantages</th>
<th>Disadvantages</th>
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<tbody>
<tr>
<td>laminectomy</td>
<td>simple, less operative time &amp; blood loss, low immediate complication</td>
<td>risk of OPLL progression; risk of kyphotic deformity, spinal instability, &amp; neurological deterioration due to scar tissue formation; ineffectiveness in cases w/ severe kyphotic deformity &amp; large OPLL</td>
</tr>
<tr>
<td>laminectomy w/ fusion</td>
<td>relatively simple, low complication rate, decreased risk of kyphotic deformity &amp; spinal instability</td>
<td>risk of OPLL progression, ineffectiveness in cases w/ severe kyphotic deformity &amp; large OPLL</td>
</tr>
<tr>
<td>laminoplasty</td>
<td>relatively simple, low complication rate compared w/ ant approach, decreased risk of kyphotic deformity, spinal instability &amp; neurological deterioration due to scar tissue formation compared w/ laminectomy alone</td>
<td>risk of OPLL progression, limited effectiveness in cases w/ severe kyphotic deformity &amp; large OPLL</td>
</tr>
<tr>
<td>ant approach</td>
<td>direct ant decompression of OPLL</td>
<td>high complication rate (particularly neurological deterioration, graft complication, &amp; CSF leakage), limitation in cases w/ long segment OPLL or OPLL involving C-2</td>
</tr>
<tr>
<td>combined ant &amp; pst approach</td>
<td>direct ant decompression of OPLL</td>
<td>more op time &amp; blood loss</td>
</tr>
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</table>

* ant = anterior; pst = posterior.

rior approach (ACDF, anterior cervical corpectomy with fusion, open-window corpectomy, oblique corpectomy, skip corpectomy, and anterior decompression via a trans-vertebral approach), and the combined anterior and posterior approach. The advantages and disadvantages of each approach are summarized in Table 2.

Posterior Approach. Laminectomy is the simplest procedure used to decompress the spinal cord from the posterior approach. Progression of kyphotic deformity after cervical laminectomy for OPLL has been reported, but its presence did not affect neurological outcomes of the patients.\(^{13,30}\) OPLL progression after laminectomy also rarely caused neurological deterioration. In the report from Kato et al.,\(^{50}\) OPLL progression was noted in 70% of patients, but it was clearly the cause of neurological deterioration in only 1 of them. A rare case\(^{36}\) of incarcerated spinal cord herniation with neurological deterioration after laminectomy in a patient with combined OPLL and OLF of the cervical spine has also been reported in the literature.

Anderson et al.\(^{4}\) found that laminectomy with fusion decreases the risk of postoperative kyphotic deformity and spinal instability compared with laminectomy alone, but functional improvement is similar to laminectomy or laminoplasty. This paper contains a description of several posterior cervical fusion techniques and the lateral mass and pedicle screws that are used by many spine surgeons. One drawback of these techniques is neurovascular injury. Hasegawa et al.\(^{39}\) reported higher operative duration and intraoperative blood loss in patients treated using pedicular screw fixation compared with those treated by laminoplasty. They concluded that there is no indication for cervical pedicular screw fixation in patients with typical OPLL and CSM because of the potential risk of vertebral artery or nerve injury. Recently, Epstein\(^{18}\) reported good outcomes with spinous process wiring techniques. Fusion rate was 100% and complications were low, including 2 transient root injuries, 2 wound infections, 1 wound breakdown, no spinal cord injuries, and no deaths.

Nerve root palsy at C-5 after cervical laminectomy and posterior fixation is correlated with increased cervical lordosis\(^{79}\) and the main pathogenic mechanism appears to be the tethering effect.

Laminoplasty has been used for decades for posterior decompression of the spinal canal in patients with cervical OPLL. The benefits of this technique compared with laminectomy are reduced risk of postoperative kyphotic deformity and neurological deficit from scar tissue formation. There are 2 techniques of laminoplasty: open-door and double-door (Fig. 4). However, both have some limitations, including restricted access to the hinged side in open-door laminoplasty, a potential for closing of the door,\(^{17}\) axial neck pain, loss of range of motion of the cervical spine, risk of OPLL progression, and limited effectiveness in cases with severe kyphotic deformity and large OPLL.

There have been some modifications of the open-door technique to prevent closing of the door, including spacer insertion at the bone gap at the open side (bone graft from spinous process or hydroxyapatite spacer),\(^{19}\) titanium miniplate fixation, and the TiMesh LP (Medtronic Sofamor Danek) miniplate system.\(^{19}\) Adequate longitudinal and transverse decompression of the spinal canal should be achieved because unexpectedly rapid progression of OPLL has been reported.\(^{122}\) Seichi et al.\(^{34}\) used intraoperative ultrasonography to evaluate adequacy of spinal cord decompression after double-door laminoplasty. They found that OPLL maximal thickness > 7.2 mm was a cutoff value for insufficient decompression, but neurological outcomes at 2 years after surgery did not correlate with adequacy of decompression.

Hirabayashi et al.\(^{33}\) compared the expansion ratio of the spinal canal and the increased inclination angle of the lamina between open-door and double-door laminoplasty. Open-door laminoplasty produced a significantly larger expansion ratio at C-6 than double-door laminoplasty. The increase of inclination angle of the lamina was significantly larger in double-door than in open-door laminoplasty. They proposed the surgical indications for open-door lami-
noplasty as CSM combined with hemilateral radiculopathy, large prominence of OPLL, and patients with a tiny spinous process who cannot undergo double-door laminoplasty. The indications for double-door laminoplasty include usual CSM, small and slight prominence of OPLL, CSM combined with bilateral radiculopathy, and cervical canal stenosis combined with instability necessitating posterior spinal instrumentation surgery.

Long-term results of open-door laminoplasty are summarized in Table 3. In studies with a long-term follow-up period (more than 5 years), the recovery rates varied from 47.9% to 63.1%.

Agrawal et al. demonstrated the benefit of expansive laminoplasty even in patients with severe cervical myelopathy (Nurick Grade 3–5). All patients with a duration of symptoms less than 3 years, and 50% of patients with durations ranging from 3 to 6 years, improved after surgery.

Factors influencing surgical outcomes following laminoplasty included duration of myelopathy, severity of myelopathy, preoperative kyphosis, occupying ratio > 60%, and hill-shaped ossification. There are controversial results of some factors such as progression of OPLL and postoperative changes in cervical alignment.

The course of neurological changes following laminoplasty has been investigated. Neurological function significantly improved after surgery, was maintained for 5 years, and then slightly declined after 5 years. Ogawa et al. found that the degree of deterioration positively correlated with cervical range of motion, which is high in patients with segmental-type OPLL.

Postoperative cervical range of motion decreased after laminoplasty by approximately 32% and did not correlate with postoperative axial neck pain. The loss of range of motion is time-dependent and plateaus by 18 months after surgery.

Anterior Approach. As mentioned above, Koyanagi et al. reported a high incidence of associated disc herniation in patients with cervical OPLL, and disc herniation was found at maximum compression level in 60% of patients. Anterior cervical disectomy with fusion is the procedure of choice for these patients. The recovery rate with this procedure ranges from 51% to 63.2%. Tan et al. reported on ACDF with an endoscopic approach in 5 patients with cervical OPLL. The patients’ JOA scores and visual analog scale scores improved significantly. This technique has advantages in terms of cosmetic results, intraoperative visualization, and recovery course, but its application is limited to the C4–5 and C5–6 levels. At the higher levels, installation of the working channel can be blocked by the mandible. At the C6–7 level, there is a risk of damaging a thyroid vessel, as reported in 1 patient in this paper. Multilevel OPLL is also a contraindication for this procedure.

Neurological improvement rates in anterior approaches to cervical OPLL are summarized in Table 4. Improvement rates varied from 51% to 71.7%. There are many varieties of techniques, bone grafts, and instruments for anterior approach surgery. Mizuno and Nakagawa used 3 graft materials for this approach including iliac crest, vertebral body, and interbody fusion cages and found that vertebral body grafts were the most fragile. Rajshekhar and Kumar performed corpectomies in poor-grade patients (Nurick Grade 4 and 5), and neuro-

![Fig. 4. Illustrations of open-door (left) and double-door (right) laminoplasty.](image)

**TABLE 3: Long-term results of open-door laminoplasty for OPLL**

<table>
<thead>
<tr>
<th>Authors &amp; Year</th>
<th>No. of Patients</th>
<th>Minimum FU (yrs)</th>
<th>Mean Neurological Recovery Rate (%) at Last FU</th>
<th>Notes</th>
</tr>
</thead>
<tbody>
<tr>
<td>Fujimura et al., 1998</td>
<td>55</td>
<td>5</td>
<td>49.3</td>
<td>duration of myelopathy was a factor indicating poor results</td>
</tr>
<tr>
<td>Iwasaki et al., 2002</td>
<td>64</td>
<td>10</td>
<td>60.0</td>
<td>progression of OPLL required additional op in 1 patient</td>
</tr>
<tr>
<td>Ogawa et al., 2004</td>
<td>72</td>
<td>5</td>
<td>63.1</td>
<td>progression of OPLL caused myelopathy in 2 patients</td>
</tr>
<tr>
<td>Chiba et al., 2006</td>
<td>80</td>
<td>10</td>
<td>47.9</td>
<td>OPLL patients w/ preoperative kyphosis had lower recovery rates</td>
</tr>
<tr>
<td>Iwasaki et al., 2007</td>
<td>66</td>
<td>5</td>
<td>58.0</td>
<td>outcome was significantly poorer in patients w/ occupying ratio &gt;60%</td>
</tr>
</tbody>
</table>

* FU = follow-up.
logical improvement was achieved in 76% of patients. They concluded that early decompressive surgery should be offered to poor-grade patients.

Onari et al. described long-term (mean 14.7 years) follow-up results of anterior interbody fusion without decompression in patients with cervical OPLL. Twenty-four of the 30 patients improved after surgery. These investigators found that this procedure was more effective for the patients with segmental or nodular type OPLL than for those with continuous or mixed type. These data indicate that a dynamic factor is an important factor contributing to myelopathy in patients with cervical OPLL.

The open window corpectomy technique has been described in the literature. This technique creates a more stable construct with 3-point fixation and offers better load sharing among implants and healthy vertebrae. Ozer et al. reported satisfactory clinical and radiological outcomes in patients with cervical OPLL after using this technique.

Oblique corpectomy preserves the ventral half of the vertebral body, so fusion and stabilization are not required. Anecdotal reports of using the oblique corpectomy technique for cervical OPLL exist in the literature, including Goel and Pareikh, who reported 4 cases successfully treated with this technique. Wide exposure for resection of OPLL was achieved and stability of the spine was also preserved. Chacko and Daniel applied this technique in 3 patients with combined OALL and OPLL. All patients showed clinical improvement. Asymptomatic OALL provided an intrinsic stability to the spine and was preserved in all patients. Intraoperative ultrasonography provided real-time imaging during surgery. Moses et al. evaluated the accuracy of intraoperative ultrasonography in patients who underwent oblique corpectomy. They concluded that it is helpful in identifying the vertebral artery and determining the trajectory of approach, but there are limitations in OPLL cases due to artifacts from residual ossification.

The skip corpectomy technique (C-4 and C-6 corpectomy with preservation of C-5 vertebral body) was reported by Dalbayrak et al. in 29 patients with multilevel CSM and cervical OPLL. The mean JOA score improved from 13.44 to 16.16 after surgery. There was only 1 case with complications from instrumentation (C-7 screw pullout). The preservation of the C-5 vertebral body improved screw purchase and strengthened the construct.

A wide transvertebral approach and a ceramic insertion for patients with cervical degenerative disease were reported by Kim et al. The advantage of this technique is preservation of the intervertebral disc, so movement of the spine is retained. The successful outcomes were achieved in patients with segmental-type OPLL. Because of the narrow visual field, this approach should not be used in patients with segmental instability, continuous or combined OPLL, and kyphosis.

Park et al. described a prevascular extraoral retropharyngeal approach to the upper cervical spine, including a case of C2–4 OPLL. They reported that this approach is relatively safe. In a series of 15 patients, there was only 1 permanent and 2 transient dysphagia cases. There were no complications related to the marginal branch of the facial nerve or submandibular gland.

**Surgical Treatment of Thoracic OPLL.** The surgical results of thoracic OPLL are poorer than those of cervical OPLL. There are several factors that limit the effectiveness of thoracic OPLL decompression. One natural kyphosis of the thoracic spine restricts the backward shift.
of the spinal cord after posterior decompression; 2) the thoracic segment of the spinal cord is relatively avascular compared with the cervical segment, therefore it is more vulnerable to ischemic injury during surgical manipulation; and 3) the ribcage restricts the surgical approaches to this area of the spine.

Surgical options for treatment of thoracic OPLL include posterior decompressive laminectomy or laminoplasty, posterior decompression and fusion, anterior decompression through an anterior approach, circumspinal decompression through a posterior approach, and 2-stage posterior and anterior decompression. Posterior decompressive laminectomy is indirect and the simplest method for thoracic OPLL decompression, but postoperative paraparesis is the main drawback of this technique. Thoracic laminectomy causes disruption of the posterior tension band of the spine, which may lead to instability and neurological deterioration. Two cases have been reported of patients with thoracic OPLL who underwent laminectomy and suffered postoperative neurological deterioration. Both patients underwent reoperation with posterior instrumented fusion and neurological functions gradually improved. The authors recommended simultaneous posterior instrumented fusion after laminectomy for thoracic OPLL. Nakanishi et al. demonstrated a case of extensive cervicothoracic OPLL in which the patient underwent thoracic laminectomy with electrophysiological monitoring of the spinal cord evoked potential. The amplitude of evoked potential decreased after laminectomy, but recovered after posterior instrumented fusion. This finding emphasizes the importance of a dynamic factor and progression of kyphosis as the causes of neurological deterioration after laminectomy. In the study of factors related to outcomes of thoracic OPLL surgery, Matsumoto et al. also recommended instrumented fusion after posterior decompression. Beak-type OPLL has higher risk of neurological deterioration after posterior approach surgery than flat-type OPLL. Beak-type and flat-type OPLL are shown in Fig. 5.

Komagata et al. studied the effectiveness of open-door laminoplasty in 13 patients with myelopathy from cervicothoracic OPLL with an average follow-up period of 75 months. According to the Hirabayashi method, the mean recovery rate was 54.5% without restenosis of the opened lamina and marked progression of kyphosis, but there were 2 cases of transient motor paralysis of both legs after the operation. A multiinstitutional study by Matsumoto et al. showed that laminoplasty can be used safely to treat thoracic OPLL at the nonkyphotic upper thoracic spine (T1–4).

Posterior decompression with fusion generally has lower complication rates and neurological deterioration compared with both posterior decompression alone and OPLL extirpation. Yamazaki et al. treated patients with thoracic OPLL by 1 of 3 approaches: posterior decompression alone (18 patients), posterior decompression and fusion (17 patients), and OPLL extirpation (16 patients). Three patients who underwent posterior decompression alone and 3 patients who underwent OPLL extirpation developed postoperative paralysis. Seven patients in the posterior decompression only group developed late neurological deterioration. There were 8 patients with CSF leakage and 2 patients with hydrothorax in the OPLL extirpation group. There were no cases of postoperative paralysis or late neurological deterioration in patients who underwent posterior decompression and fusion. Recovery of neurological function after posterior decompression and fusion is another challenge because the natural curve of the thoracic spine is kyphosis and posterior decompression may be ineffective. Yamazaki et al. performed posterior decompression with in situ instrumented fusion in 24 patients with thoracic OPLL. The mean follow-up period was 4 years and 5 months, the mean recovery rate was 58.1%, and the median time to point of maximal recovery was 9 months. Only 1 patient developed transient paralysis. Despite persistent impingement of the spinal cord by OPLL, considerable neurological improvement is expected with this technique and takes about 9 months before reaching maximal recovery.
Ossification of the posterior longitudinal ligament

Posterior decompression with kyphosis correction has been studied. The recovery rate varied from 56% to 68%.79,143 Zhang et al.144 performed posterior decompression with 5°–15° kyphosis correction with instrumented fusion in 11 patients. Postoperative MR imaging showed backward shift of the spinal cord and complete decompression in all cases without aggravated myelopathy. Matsuyama et al.29 used intraoperative ultrasonography to evaluate backward shift of the spinal cord and intraoperative compound muscle action potential to determine correlation with the final outcomes. There was no significant difference in recovery rate between adequate and inadequate decompression detected by ultrasonography, but patients with decreased compound muscle action potential had significantly poorer recovery rates.

Tokuhashi et al.127 tried to determine the critical ossification-kyphosis angle that affects surgical outcome in patients who underwent posterior decompression of thoracic OPLL. At the decompression site, 23° is the critical cut-off point. All patients with an ossification-kyphosis angle > 23° had no echo-free space detected by intraoperative ultrasonography, whereas all patients with < 23° had echo-free space.

The anterior approach to OPLL resection has the benefit of direct OPLL removal, but it is technically demanding and the surgical results are poor, particularly in patients who already had severe spinal cord compression before surgery. In 2008, Min et al.82 reported high rates of complications of anterior approach decompression in 19 patients with thoracic OPLL. Two patients (10.5%) developed neurological deterioration and 6 patients (31.6%) developed CSF leakage. They also demonstrated that poor outcomes of this approach were associated with poor preoperative JOA scores and immediate postoperative neurological deterioration.

The largest benefit of circumspinal decompression through the posterior approach is immediate anterior and posterior decompression and/or stabilization with only 1 operation. Yang et al.129 reported satisfactory outcomes in a case with T10–11 OPLL surgically treated using this technique. Takahata et al.118 reported on 30 patients who underwent this type of surgery with a mean follow-up period of 8 years. The JOA score improved in 24 patients (80%). Surgical complications included 40% with a dural tear, 10% with a deep infection, and 33% with postoperative neurological deterioration. Patients who underwent decompression of 5 or more vertebral levels had poorer outcomes.

Anatomical factors inhibiting posterior shift of the spinal cord after posterior decompression were described by Tsuzuki et al.127 Longitudinal factors are anterior pulling effects of spinal cord segments above and below OPLL and restraining effects of dural dura. These factors can be eliminated by extensive cervicothoracic laminoplasty and or without posterior longitudinal durotomy. Axial factors are anterior adhesion of dura to OPLL, dural ossification, and an anterior tethering effect of thoracic roots and dentate ligaments. These factors can be eliminated by root release with total laminofacetectomy and anterolateral dural release with or without OPLL resection. These investigators used staged posterior approach surgery to address these problems. The advantage of this staged operation is its safety, by preparing the severely compressed spinal cord by a first-stage operation before undergoing extensive manipulation by a second-stage surgery. The first stage consisted of extensive cervicothoracic laminoplasty decompression with or without posterior longitudinal durotomy to eliminate the longitudinal factors. If the decompression was inadequate, the axial factors were eliminated by the above mentioned techniques. In their series of 17 patients with a mean follow-up period of 42 months, neurological improvements were comparable to those from a successful anterior approach decompression. Only 1 case of late neurological deterioration was encountered, caused by an arachnoid cyst compressing the dorsal spinal cord.

A case report of circumspinal decompression was presented by Hioki et al.52 Their case involved a woman with OPLL extending from C-3 to T-2 and OLF at T-2. She presented with paraparesis and numbness in both legs. After C3–T1 laminoplasty and T2–3 laminectomy, her neurological symptoms improved immediately. However, symptoms recurred after sitting or standing. A second operation was performed by anterior decompression, which improved her symptoms. Spinal instability or progression of kyphosis might have been the cause of neurological deterioration after the first surgery. Kawahara et al.53 reported on a series of 11 patients who underwent circumspinal decompression with dekyphosis stabilization. The mean JOA score improved from 4.0 to 9.1 after the operation. There were 3 patients with CSF leakage and 1 patient with postoperative neurological deterioration due to spinal cord compression by the swelling of paravertebral muscle.

Surgical outcomes of patients with thoracic myelopathy were correlated with preoperative duration of symptoms and degree of myelopathy. Patients with shorter duration of symptoms and milder myelopathy experienced better surgical outcomes.3 To date, there are no definitive guidelines for surgical treatment of thoracic OPLL. The choice of operation should be selected on a case-by-case basis, depending on the patient's condition, level of pathology, type of OPLL, and experience of the surgeon. Advantages and disadvantages of surgical procedures for thoracic OPLL are summarized in Table 5.

Surgical Management of Lumbar OPLL. There are some reports of surgical treatment of lumbar OPLL, but the definitive procedure has not been established. Most of the cases were approached posteriorly. Symptomatic lumbar OPLL is usually located at the upper lumbar spine because the posterior longitudinal ligament is broader at the upper level. Patients may present with cauda equina syndrome. Tamura et al.117 reported on a patient with lumbar OPLL who underwent an operation using the anterior approach and another patient who underwent a combined anterior-posterior approach. The authors recommended combined surgery in patients with OPLLs occupying large parts of the spinal canal.

Conclusions

OPLL is a common cause of myelopathy in Asian
populations. While the pathogenesis of this disease is still unclear, genetic, hormonal, environmental, and lifestyle factors are believed to cause OPLL formation and progression. Occurrence of myelopathy in patients with OPLL is related to both static and dynamic factors. Radiological evaluation of OPLL includes plain radiography, CT, and MR imaging. Preoperative images should be meticulously evaluated to detect the maximal area of spinal cord compression and dural calcification, which is rather accurately demonstrated by a double-layer sign on CT scans. Surgical management of OPLL remains controversial; each approach has its own limitations, advantages, and disadvantages. The choice of operation should be made on a case by case basis, depending on the patient’s condition, level of pathology, and type of OPLL, as well as the experience of the surgeon. Most published papers in the literature are case series and retrospective studies, but more prospective studies and improvement of genetic studies will be key to more thoroughly understanding the pathogenesis, OPLL progression, myelopathy progression, and optimal treatment of patients with OPLL.

Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

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References


TABLE 5: Summary of advantages and disadvantages of surgical procedures for thoracic OPLL

<table>
<thead>
<tr>
<th>Surgical Procedure</th>
<th>Advantages</th>
<th>Disadvantages</th>
</tr>
</thead>
<tbody>
<tr>
<td>pst decompression</td>
<td>simple, less op time &amp; blood loss</td>
<td>high risk for postop paralysis &amp; late neurological deterioration</td>
</tr>
<tr>
<td>pst decompression w/ fusion</td>
<td>less op time &amp; blood loss compared w/ ant or combined approach, low risk of postop paralysis</td>
<td>persistent ant impingement of spinal cord by OPLL</td>
</tr>
<tr>
<td>ant decompression through ant approach</td>
<td>direct removal of OPLL</td>
<td>high risk for postop paralysis &amp; CSF leakage, technically demanding, more op time &amp; blood loss</td>
</tr>
<tr>
<td>spinal decompression through pst approach</td>
<td>immediate ant &amp; pst decompression &amp; stabilization w/ only 1 op</td>
<td>technically demanding, more op time &amp; blood loss</td>
</tr>
<tr>
<td>2-stage pst &amp; ant decompression</td>
<td>complete ant &amp; pst decompression</td>
<td>technically demanding, more op time &amp; blood loss</td>
</tr>
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K. Saetia et al.
Ossification of the posterior longitudinal ligament


Ossification of the posterior longitudinal ligament


