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Analysis of demographics, risk factors, clinical presentation, and surgical treatment modalities for the ossified posterior longitudinal ligament

Samuel Kalb, M.D.,[1] Nikolay L. Martirosyan, M.D.,[1] Luis Perez-Orribo, M.D.,[2] M. Yashar S. Kalani, M.D., Ph.D.,[1] and Nicholas Theodore, M.D.[1]

[1]Division of Neurological Surgery, Barrow Neurological Institute, St. Joseph's Hospital and Medical Center, Phoenix, Arizona; and [2]Division of Neurosurgery, Hospital Universitario de Canarias, Santa Cruz de Tenerife, Canarias, Spain

Object. Ossification of the posterior longitudinal ligament (OPLL) is a rare disease that results in progressive myeloradiculopathy related to pathological ossification of the ligament from unknown causes. Although it has long been considered a disease of Asian origin, this disorder is increasingly being recognized in European and North American populations. Herein the authors present demographic, radiographic, and comorbidity data from white patients with diagnosed OPLL as well as the outcomes of surgically treated patients.

Methods. Between 1999 and 2010, OPLL was diagnosed in 36 white patients at Barrow Neurological Institute. Patients were divided into 2 groups: a group of 33 patients with cervical OPLL and a group of 3 patients with thoracic or lumbar OPLL. Fifteen of these patients who had received operative treatment were analyzed separately. Imaging analysis focused on signal changes in the spinal cord, mass occupying ratio, signs of dural penetration, spinal levels involved, and subtype of OPLL. Surgical techniques included anterior cervical decompression and fusion with corpectomy, posterior laminectomy with fusion, posterior open-door laminoplasty, and anterior corpectomy combined with posterior laminectomy and fusion. Comorbidities, cigarette smoking, and previous spine surgeries were considered. Neurological function was assessed using a modified Japanese Orthopaedic Association Scale (mJOAS).

Results. A high-intensity signal on T2-weighted MR imaging and a history of cervical spine surgery correlated with worse mJOAS scores. Furthermore, mJOAS scores decreased as the occupying rate of the OPLL mass in the spinal canal increased. On radiographic analysis, the proportion of signs of dural penetration correlated with the OPLL subtype. A high mass occupying ratio of the OPLL was directly associated with the presence of dural penetration and high-intensity signal. In the surgical group, the rate of neurological improvement associated with an anterior approach was 58% compared with 31% for a posterior laminectomy. No complications were associated with any of the 4 types of surgical procedures. In 3 cases, symptoms had worsened at the last follow-up, with only a single case of disease progression. Laminoplasty was the only technique associated with a worse clinical outcome. There were no statistical differences ($p > 0.05$) between the type of surgical procedure or radiographic presentation and postoperative outcome. There was also no difference between the choice of surgical procedure performed and the number of spinal levels involved with OPLL.

Conclusions. Ossification of the posterior longitudinal ligament can no longer be viewed as a disease of the Asian population exclusively. Since OPLL among white populations is being diagnosed more frequently, surgeons must be aware of the most appropriate surgical option. The outcomes of the various surgical treatments among the different populations with OPLL appear similar. Compared with other procedures, however, anterior decompression led to the best neurological outcomes. *(DOI: 10.3171/2010.12.FOCUS10265)*

Key Words • ossification of the posterior longitudinal ligament • laminoplasty • laminectomy • anterior cervical decompression • modified Japanese Orthopaedic Association Scale

Ossification of the posterior longitudinal ligament is a pathological ectopic ossification of this ligament that usually occurs in the cervical or thoracic spine and, less frequently, in the lumbar spine.[37] It has long been considered a disease of Asian origin, and more specifically of the Japanese population. Increasingly, however, this disorder is being recognized in European and North American populations. The prevalence of OPLL in Japanese and East Asian countries has ranged from 1.9% to 4.3%,[17,21,34,42,43] while in white populations it has ranged from 0.01% to 1.7%.[14,21]

The typical age at onset is 50 years, with a male/female ratio of 2:1.

The Japanese Investigation Committee on the Ossification of the Spinal Ligaments[42,43] defined 4 subtypes of OPLL based on the extent of the condition: 1) focal, local-

Abbreviations used in this paper: ACD = anterior cervical decompression; DISH = diffuse idiopathic skeletal hyperostosis; mJOAS = modified Japanese Orthopaedic Association Scale; OLF = ossification of the ligamentum flavum; OPLL = ossification of the posterior longitudinal ligament.

ized to the disc space; 2) segmental, located behind each vertebral body and not extending beyond the adjacent disc level; 3) continuous, between several levels overpassing the disc; and 4) mixed, combination of segmental and continuous types. The segmental type occurs in most cases (39%) followed by the mixed (29%), continuous (27%), and focal types (5%).[3]

Clinical presentations correspond to the level and magnitude of spinal cord compression. Cervical and thoracic OPLL typically manifests with signs and symptoms of myelopathy, while the lumbar disease usually manifests with signs and symptoms of stenosis.[17,27,29] Predictors and risk factors for the development or progression of myelopathy include > 60% OPLL-induced spinal stenosis, increased range of motion of the cervical spine, progression of OPLL, type of OPLL (that is, the segmental type is associated with the greatest risk), and lateral-deviated OPLL.[25–27,30] After 30 years, however, myelopathy-free rates as high as 71% have been reported among patients who had no myelopathy when the OPLL was first diagnosed.[27]

Surgical procedures for the treatment of cervical OPLL can be separated into anterior or posterior approaches. The anterior approach involves direct removal of the ossified mass, while the posterior approach involves laminoplasty, laminectomy, or laminectomy with posterior fusion for decompression and stabilization. Rates of neurological improvement after anterior surgery have been as high as 92% and have been associated with fewer complications than the posterior procedures.[27,29,40]

Both laminectomy and laminoplasty are safe and effective treatments for high-risk patients with multilevel OPLL. The rates of neurological improvement associated with laminoplasty and laminectomy have been reported to be 67% and 42%, respectively.[12,15] However, significant neurological deterioration immediately after surgery, cervical kyphosis, and progression of ossification are concerns when a using these techniques.[12,16,24,40]

Several factors have been associated with negative surgical outcomes, including age, misalignment of the cervical spine, preoperative neurological score, intramedullary high-intensity signal on sagittal T2-weighted MR imaging, OPLL subtype, and a mass occupying ratio ≥ 60%.[12,22,24,36,40,45]

As OPLL is recognized and its incidence increases accordingly, it is worthwhile to understand trends related to this disease. We therefore analyzed the demographics, clinical and imaging features, and potential risk factors associated with OPLL in a population of white patients. Note that the surgical experience in white patients with OPLL is limited as compared with that in the Asian population; consequently, we evaluated our institutional experience in these patients.

Methods

This study was approved by the Institutional Review Board of St. Joseph's Hospital and Medical Center in Phoenix, Arizona.

Between September 1999 and September 2010, symptomatic OPLL was diagnosed in 41 white patients at the Barrow Neurological Institute, and data were retrospectively reviewed in 36 of them for this study. Data for the remaining 5 patients were not sufficient for analysis. Sixteen patients (44.5%) were males, and 20 (55.5%) were females, with a mean age of 56 years (range 32–85 years). Patients were divided into 2 groups based on the anatomical location of their OPLL: a group of 33 patients (16 males [48.5%] and 17 females [51.5%], with a mean age 56 years) with cervical OPLL and a group of 3 female patients (mean age 49 years) with thoracic (2 patients) or lumbar (1 patient) OPLL.

Analyzed comorbid conditions included diabetes mellitus, systemic hypertension, dyslipidemia, hypothyroidism, meningioma, DISH, cigarette smoking, and previous lumbar surgeries (Table 1). Each patient's family history for disease as well as any history of trauma was documented. Analyzed imaging data included high-intensity signal of the spinal cord on T2-weighted MR imaging, mass occupying ratio (percentage of spinal canal diameter occupied by the OPLL) on CT scanning, type of OPLL, signs of dural penetration as described by Hida et al.,[8] and involved spinal levels.

Of the patients identified, 15 were surgically treated (Table 2). Ten (66.7%) were males and 5 (33.3%) were females, with a mean age of 56 years (range 32–75 years). The mean duration of follow-up among this group was 22.5 months (range 1–150 months). Cervical OPLL was present in 14 cases. Thoracic OPLL was present in 1 case, which was separately evaluated because of the difference in its clinical presentation and management. Inclusion into this small surgical series required a patient to have undergone OPLL surgery performed by a senior neurosurgeon at the Barrow Neurological Institute. Surgically treated patients with OPLL were excluded if their clinical presentation or indication for spine surgery was unrelated to OPLL; if there were no data on the patient's demographics, comorbid conditions, and follow-up; or if no appropriate diagnostic imaging studies were available after a thorough review of the patient's medical records, computerized data, films, and charts.

Patients underwent the following surgical approaches (Table 3): ACD and fusion with corpectomy, posterior laminectomy with fusion, posterior open-door laminoplasty, and combined anterior corpectomy with posterior laminectomy and fusion (360° approach). The clinical pre-

TABLE 1: Comorbid conditions in 36 white patients with OPLL

Condition	No. (%)	
	Cervical OPLL	Thoracic-Lumbar OPLL
diabetes mellitus	11 (33.3)	1 (33.3)
systemic hypertension	19 (57.6)	2 (66.7)
dyslipidemia	4 (12.1)	
DISH	1 (3.0)	
hypothyroidism	3 (9.1)	
meningioma	3 (9.1)	1 (33.3)
cigarette smoking	5 (15.2)	
history of trauma	9 (27.3)	
history of spine op	3 (9.1)	

TABLE 2: Summary of clinical characteristics in 15 patients who underwent surgical treatment for OPLL*

Case No.	Age (yrs), Sex	Comorbidity/Surgical or Medical History	Level of Disease	Type of Disease†	Level of Op	Type of Op
1	65, F	systemic hypertension, dyslipidemia, cervical spine op	C2–6	continuous	C2–7	laminectomy & fusion
2	51, M	diabetes mellitus, systemic hypertension, DISH	C2–T1	continuous	C4–7	360°
3	32, F	none	C3–5	segmental	C3–6	ACD & fusion w/ corpectomy
4	49, F	systemic hypertension, meningioma	C3–6	continuous	C3–6	laminectomy & fusion
5	75, M	systemic hypertension, dyslipidemia, diabetes mellitus	C5–7	segmental	C5–T1	360°
6	56, M	systemic hypertension	C4–5	segmental	C3–6	laminoplasty
7	47, M	systemic hypertension	C3–7	mixed	C3–7	laminectomy & fusion
8	73, M	systemic hypertension	C3–6	segmental	C3–7	ACD & fusion w/ corpectomy
9	45, M	none	C4–7	Segmental	C4–7	ACD & fusion w/ corpectomy
10	73, M	none	C2–4	mixed	C2–5	laminectomy & fusion
11	63, M	none	C2–4	mixed	C2–5	laminectomy & fusion
12	53, F	systemic hypertension, dyslipidemia, diabetes mellitus, hypothyroidism	C1–5, C7–T1	mixed	C1–4	laminectomy & fusion
13	53, M	systemic hypertension, smoking	C3–T1	mixed	C3–6	laminectomy & fusion
14	58, M	none	C4–6	mixed	C3–6	laminectomy & fusion
15	32, F	systemic hypertension	T3–10, T-12	mixed	T6–10	laminectomy & fusion

* 360° = ACD and corpectomy with posterior laminectomy and fusion.

† Type of OPLL as classified by the Japanese Investigation Committee on the Ossification of the Spinal Ligaments. See Tsuyama, 1984.

sentation associated with OPLL and treatment outcomes were assessed using the mJOAS as described by Benzel et al.,[1] in which motor, sensitivity, and sphincter functions were scored. The maximum mJOAS score, corresponding to normal, for patients with cervical OPLL was 18, and the maximum score for patients with thoracic or lumbar OPLL, which excludes upper limb function, was 13. Subsequently, the recovery rate was calculated using the Hirabayashi et al.[9] method (recovery rate = [postoperative mJOAS score – preoperative mJOAS score] × 100/(maximum score – preoperative mJOAS score) and was defined as excellent (100%–75%), good (74%–50%), fair (49%–25%), unchanged (24%–0%), or deteriorated (score < 0%). Intraoperative and postoperative (follow-up) complications were also analyzed.

Statistical analysis was performed with a personal computer operating SPSS, version 18. Independent t-tests were used to analyze demographic and comorbid conditions by radiographic presentation. A post hoc test (Fisher least significant difference) was applied to determine differences in radiographic features at presentation (that is, mass occupying ratio and dural penetration) by type of OPLL. Analysis of variance was utilized to compare mean scores by OPLL type and surgery type as well as to analyze outcome variables by the type of surgery performed. A p value ≤ 0.05 was considered significant.

Results

Cervical OPLL

Patients with cervical OPLL (Table 4) had the following OPLL types: localized, 9 patients (27.3%); continuous, 5 patients (15.2%); segmental, 12 patients (36.4%); and mixed, 7 patients (21.2%). One-level disease was present in 4 cases (12.1%), 2-level disease in 9 (27.3%), 3-level disease in 9 (27.3%), 4-level disease in 3 (9.1%), 5-level disease in 5 (15.2%), and 6-level disease in 3 (9.1%). Signs of dural penetration were evident in 16 patients (48.5%), whereas high-intensity T2-weighted MR imaging signals were present on the spinal cord in 17 cases (51.5%). On axial CT scans, the mean mass occupying ratio was 38.7%

TABLE 3: Preoperative and postoperative neurological status of 15 patients based on the mJOAS as a function of surgical approach

Op Type	No. of Cases	Mean Preop mJOAS Score	Mean Postop mJOAS Score	Mean Recovery Rate (%)*	Recovery Rate Result*
laminectomy	9	11.2	14.1	36.5	fair
laminoplasty	1	14.0	12.0	−50.0	deteriorated
ACD & corpectomy	3	15.3	17.0	58.3	good
360°	2	12.0	13.5	31.2	fair

* According to Hirabayashi method: recovery rate = (postoperative mJOAS score − preoperative mJOAS score) × 100/(maximum score − preoperative mJOAS score).

TABLE 4: Cases of OPLL by cervical level

Level	No. (%)
C-1	1 (3.0)
C-2	9 (27.3)
C-3	14 (42.4)
C-4	22 (66.7)
C-5	23 (69.7)
C-6	23 (69.7)
C-7	10 (30.3)

(range 16%–74.5%). The mean score on the mJOAS, as a reflection of clinical status, was 14.3 (range 8–18). Figure 1 shows the average mJOAS score for each comorbid and radiographic condition.

The mJOAS scores were significantly worse for patients whose T2-weighted MR imaging studies showed a high-intensity signal from the spinal cord ($p = 0.001$, 12.8 vs 16 in those without T2-weighted signal from the spinal cord) and for patients with a history of cervical spine surgery unrelated to OPLL ($p = 0.003$, 14.3 vs 16 in those without previous spine surgery). Furthermore, mJOAS scores correlated negatively with the mass occupying rate of the OPLL mass in the spinal canal ($p = 0.001$). Patients with an occupying rate > 50% had a mean mJOAS score of 11.6, as compared with a score of 14.9 in patients with a rate < 50% (no patients had a 50% mass occupying rate).

There was a significant difference in the likelihood of signs of dural penetration based on OPLL type ($p = 0.007$); that is, the mixed type of OPLL had less dural involvement than the localized ($p = 0.005$) and segmental types ($p = 0.001$). Furthermore, there were significant differences in the mean occupying ratio in the spinal canal by OPLL type ($p = 0.028$). Post hoc analysis showed that the mixed type of OPLL had a significantly higher occupying ratio than the localized ($p = 0.011$) or segmental types ($p = 0.008$).

Signs of dural penetration on CT or of high-intensity signal on T2-weighted MR images of the spinal cord were directly associated with the mass occupying ratio ($p = 0.005$ and $p = 0.001$, respectively); that is, a higher occupying ratio (mean 45% ± 13%) was directly correlated with the presence of either radiographic sign.

Thoracic and Lumbar OPLL

Radiographic evaluation showed the following involvement of the OPLL in 3 patients: a single level at T-9, 9 levels at T3–10 and T-12, and 4 levels at L1–4. Of the thoracic cases, 1 was localized and 1 was mixed, whereas the single lumbar case was a segmental type. One of the thoracic cases (whose mass occupying ratio was 73.3% on axial CT) had signs of dural penetration on CT and a high-intensity signal on T2-weighted MR images of the spinal cord. The patient in this case had an mJOAS score of 8 of 13 (after adjusting the scale without considering upper limb function), as compared with a score of 11 in the second case of thoracic OPLL, which lacked signs of both dural penetration and high-intensity signal on T2-weighted MR imaging. The sole lumbar case showed radiographic signs of dural penetration, had a mass occupying ratio of 40.5%, and an mJOAS score of 13.

Surgically Treated Group

No surgical complications occurred. At the last follow-up, however, symptoms had worsened in 3 patients. One patient had undergone a combined anterior corpectomy and posterior laminectomy and fusion (360°), and yet disease had progressed. A second patient had undergone posterior laminectomy and fusion and presented with a newly diagnosed cervical syrinx. A third patient had undergone laminoplasty, and his status had worsened for unknown reasons. Based on radiographic evaluation, the subtypes of cervical OPLL were as follows: none were localized, 3 (20%) were continuous, 5 (33.3%) were segmental, and 7 (46.7%) were mixed. Signs of dural penetration were evident in 9 cases (60%). Sagittal T2-weighted MR images showed high-intensity signal

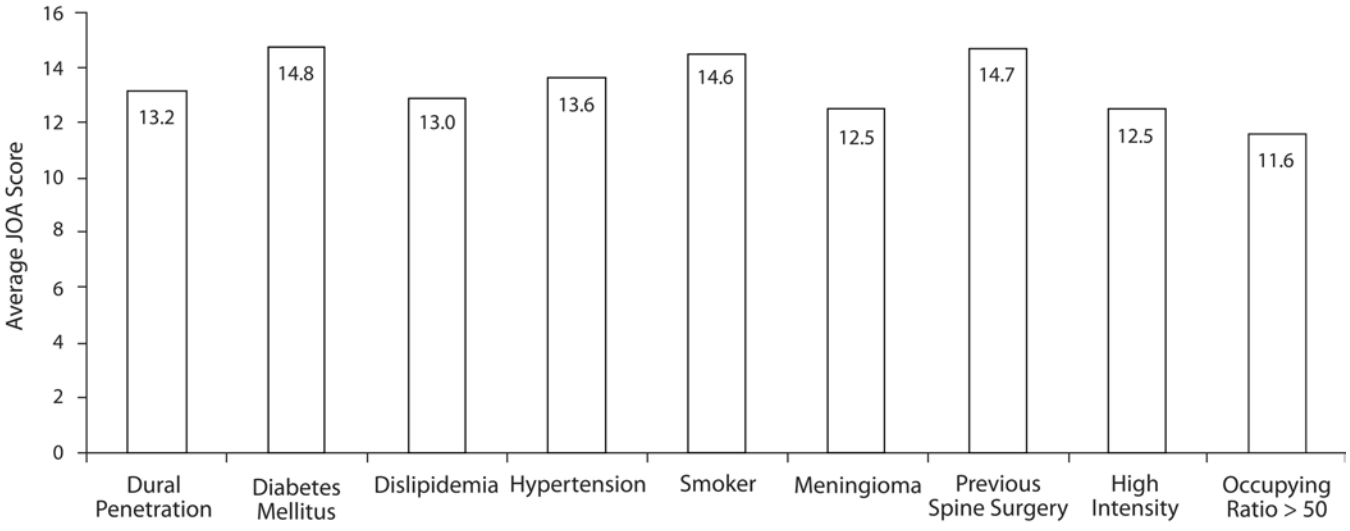

Fig. 1. Bar graph depicting mJOAS scores, measures of neurological function, as a function of demographic features and radiographic presentation.

in 11 cases (73.3%). The mean occupying ratio was 46.6% (range 24%–74.5%).

Overall, the mean improvement in mJOAS scores after all surgical procedures was 1.85. Individual mJOAS scores according to the type of surgery are shown in Figs. 2 and 3. At the last follow-up, laminoplasty was the only surgical technique associated with a worsening clinical outcome (Table 3). Comparisons of the type of surgical procedure or radiographic presentation with postoperative outcome (mJOAS score improvement and recovery rate) failed to reach significance ($p > 0.05$). Furthermore, no difference was found between the choice of surgical procedure and the number of levels involved by the OPLL (mean number of levels 4.29)

The only patient with thoracic OPLL who underwent surgical treatment was a 32-year-old woman with a mixed type OPLL involving T3–10 and T-12. She underwent laminectomy and fusion from T-6 to T-10 without intraoperative or follow-up complications. Her preoperative radiographic evaluation demonstrated an occupying ratio of 73.3% of the spinal canal space with signs of dural penetration and high-intensity signal on sagittal T2-weighted MR imaging. Her preoperative mJOAS score of 8 improved to 12 (after adjusting the scale without considering upper limb function), an 80% recovery rate.

Discussion

With an estimated rate of 0.12% in North America, the prevalence of OPLL is much lower in the white population than in the Asian population.[35] However, its prevalence is likely to increase as awareness of the disease grows among spine surgeons. Most cases in North America are reported as sporadic; however, in vitro characteristics of cell lines from American patients with OPLL are similar to those from East Asian patients,[5] which can account for the similarity of our results with findings in previously published studies in Asian-based patients with OPLL in which specific comorbidities and radiographic markers correlated with the clinical neurological presentation of disease.

It has been estimated that 70% of OPLL cases involve the cervical spine, 15% the thoracic spine, and 15% the upper lumbar spine (L1–3). The disease appears to have a multifactorial etiology in which genetic and environmental components interact. Recent studies indicate that single nucleotide polymorphism in the collagen 11A2 gene (*COL11A*) located within the Class II histocompatibility complex region on chromosome 6, which encodes the α2 chain of the Type XI collagen, could be responsible.[20,21] Two single nucleotide polymorphisms at intron 6 and exon 6 of *COL11A* could be helpful markers of this disease, and the latter could explain the sex difference in the prevalence of OPLL, which has a male/female ratio of 2:1.[23,36]

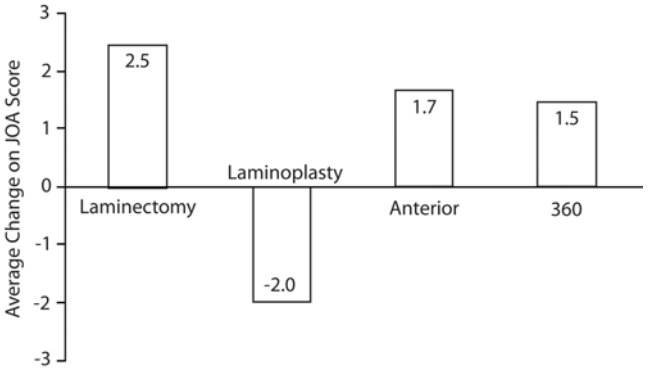

Fig. 2. Bar graph demonstrating changes in neurological status as measured by the mJOAS, as a function of surgical treatment.

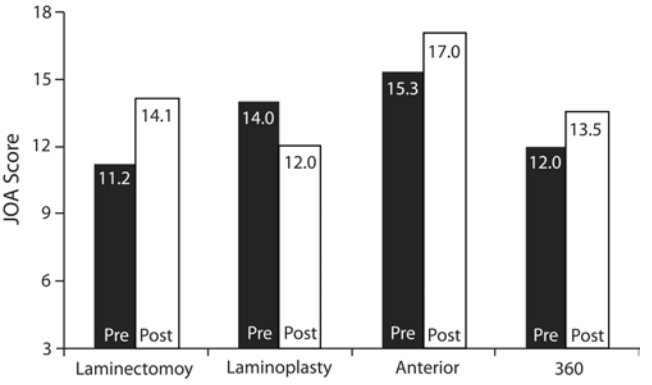

Fig. 3. Bar graph showing a comparison of preoperative and postoperative neurological status as measured by the mJOAS, as a function of surgical technique. 360 = 360° procedure.

The prevalence of polymorphisms at the collagen 6A1 gene (*COL6A1*), located on chromosome 21q, which encodes the α1 chain of Type VI collagen, is significantly greater in patients with OPLL than in healthy controls.[39] Several authors have suggested that the overproduction of both Type VI and XI collagen, as a result of the genetic alteration on their encoding genes, provides a framework for osteoblasts and/or chondrocytes to generate ectopic endochondral ossification.[20,23,39] However, the qualitative abnormality of these collagen molecules in patients with OPLL has not been demonstrated.[10]

Imaging of the spine continues to be the most appropriate diagnostic tool (Fig. 4). Lateral plain radiographs are used to measure the percentage of spinal canal occupied by the OPLL. Transaxial CT scans are the best means of visualizing the diseased ligament by showing sclerotic bone extending from the posterior side of the vertebral body into the spinal canal.[28] A sagittal overview CT is best for assessing the actual extent and subtype of OPLL and is the image of choice for evaluating dural penetrance according to the single- or double-layer sign. And T2-weighted MR imaging is essential for evaluating spinal cord swelling, myelomalacia, or gliosis within the spinal cord.[3] As diagnostic tools, additional complementary tests, including those measuring bone mineral density and bone formation markers, have been associated with promising results.[10,38]

Numerous medical factors, such as hypoparathyroidism, hypophosphatemic rickets, spondyloepiphyseal dysplasia, myotonic muscular dystrophy, obesity, DISH, ankylosing spondylitis, OLF, and a high salt/low animal protein diet, have already been identified as potential risk or associated factors for OPLL.[3,7,10,19,31,36,44] Kobashi et al.[19] reported a higher frequency of diabetes mellitus among

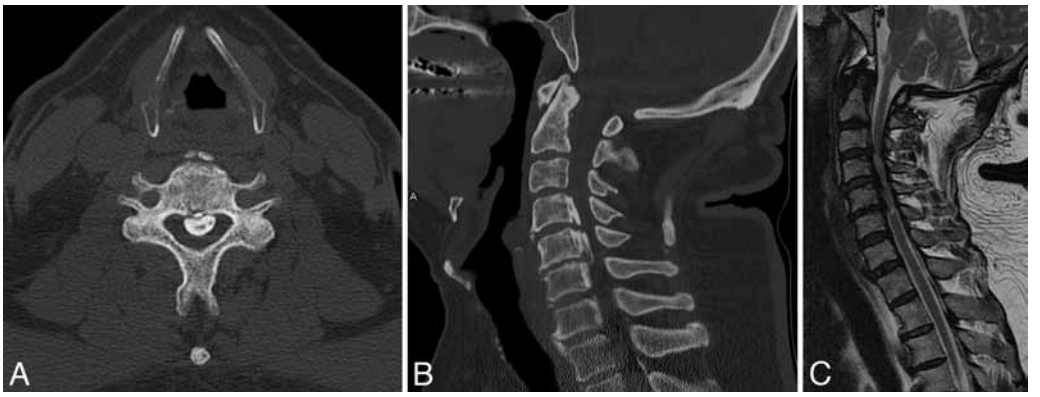

Fig. 4. Representative images obtained in a 58-year-old man with cervical OPLL from C-4 to C-6. Axial CT scan **(A)** and sagittal CT reconstruction **(B)** demonstrating an OPLL mass. Sagittal T2-weighted MR image **(C)** showing high signal intensity in the medullary region from C-4 to C-6.

Japanese men and women with OPLL as compared with controls (20% vs 5% in men, 27.6% vs 1.7% in women). In a Japanese study conducted by Ehara et al.,[2] the rate of concurrent DISH and OPLL reached 25%. In that same study the rate of concurrent OPLL and OLF was 16.7%. In a North American population, McAfee et al.[28] found that the rate of concurrent DISH and OPLL was 50%, concurrent ankylosing spondylitis and OPLL was 2%, and concurrent OLF and OPLL was 6.8%. Similarly, Epstein[4] found that OLF was present in 20% of the OPLL cases she analyzed.

In the present study, we found comorbid conditions previously reported as potential risk factors in both Asian and non-Asian populations. The prevalence of diabetes mellitus was 34.4% in those with cervical OPLL and 33.3% in those with thoracic or lumbar OPLL. One patient also had DISH. Note, however, that the prevalence of systemic hypertension was extremely high among both groups (cervical 57.5%, thoracic-lumbar 66.7%). Hypothyroidism (9.4%), dyslipidemia (12.1%), and a history of meningioma (11.1%) were also notable.

Because our population was entirely white, we used the mJOAS to measure neurological function instead of the original scale. In our attempt to identify risk factors correlating with a neurological manifestation of OPLL, we found that patients with high-intensity signal on T2-weighted MR imaging studies of the spinal cord had significantly worse mJOAS scores. These results are consistent with those of Yagi et al.,[45] who found that preoperative JOAS scores were lower in patients whose MR images showed changes in signal intensity than in those whose did not (8.8 ± 1.1 compared with 10.2 ± 1.3, respectively). Furthermore, mJOAS scores decreased as the occupying rate of the OPLL mass in the spinal canal increased. This phenomenon was also reported by Matsunaga et al.,[26] who found that patients with ≥ 60% canal stenosis also had myelopathy.

Dural penetration in diagnosed OPLL cases represents a challenge when surgical treatment is considered because of the risk of creating a dural defect and a subsequent CSF leak when the mass is removed. Consequently, CT films must be carefully evaluated to identify dural involvement indicated by single- or double-layer signs.[8] Furthermore, according to our results and those of Hida et al.,[8] a high mass occupying ratio is likely to be associated with dural penetration.

To determine the most appropriate treatment, several conditions must be regarded. Conservative management should be considered when neurological signs or symptoms (mainly neck or arm pain) are minimal and there is no evidence of myelopathy. Immobilization using cervical orthoses and skull traction combined with steroidal or nonsteroidal pharmacological agents is the available option for conservative treatment.[3,13]

Because long-lasting spinal cord compression can cause irreversible damage, surgical decompression is indicated for cervical OPLL when myelopathy is noticeable. However, the optimal surgical approach is controversial.[27] Anterior corpectomy with resection of the ossified mass followed by fusion is a radical surgical procedure best indicated for a local or segmental type of OPLL that extends fewer than 3 vertebral levels below C-2 and above T-1 in a patient with no congenital stenosis. The ossified mass should be hill-shaped, the occupying ratio should be ≥ 60%, and local kyphosis of the spinal cord should be present.[13,27,29] In contrast, the posterior surgical approach, mainly laminoplasty, is widely used to treat high-risk patients older than 65 years with multilevel disease and a nonkyphotic deformity.[3]

Even though the anterior approach for cervical OPLL is more technically demanding than a posterior approach, the best rates of improvement and functional neurological outcomes have been associated with the former. Tani et al.[40] reported a 58% rate of improvement in patients undergoing anterior decompression as compared with a 13% improvement rate in those undergoing posterior laminoplasty. In this same study, significant neurological deterioration occurred in 5 patients who had undergone laminoplasty, immediately after surgery in 4 and during late follow-up in 1. Similarly, Iwasaki et al.[11] reported a 5% rate of immediate and a 16% rate of late neurological deterioration after laminoplasty. Late-onset neurological decline is compatible with reports of postoperative progression of OPLL in patients who undergo laminoplasty. Although the frequency of OPLL progression after laminoplasty has been reported to be as high as 70%,[11,15] rates

of its symptomatic progression are low.[15,32] Additional surgical complications, primarily associated with the anterior approach, include CSF leakage and bone graft extrusions or pseudarthrosis.

In our study, patients who underwent ACD and corpectomy with fusion had the best outcomes and no surgical complications. The improvement rate following the anterior approach was 58% compared with 28.2% following the posterior approach. These results are consistent with those in previous reports.[12,40] Furthermore, as previously shown,[14,23] laminoplasty is associated with the worst outcomes, although we performed this procedure in only 1 case. Disease progression was evident in a single case—a patient who underwent combined anterior and posterior surgery. This patient had the longest follow-up (150 months). Due to the gradual nature of the disease, it is possible that in other cases progression would be apparent on longer follow-up.

In contrast to previous indications for choosing the surgical approach, the number of involved levels was not a criterion in the choice of surgery. The mean number of spinal levels involved with OPLL was approximately 4, and the outcomes of all techniques were promising. Furthermore, if laminoplasty is excluded (1 case with a negative outcome), there was no significant difference between the procedure performed and neurological outcome ($p = 0.206$) or improvement score ($p = 0.372$). Hence, neurological function improved after all procedures.

In our series, the primary indication for surgical treatment was the presence of deteriorating myelopathic symptoms with or without its associated radiographic presentation. Most of the cases showed spinal changes in the spinal cord on MR imaging and a high mass occupying ratio. Excluding laminoplasty, all patients' overall neurological function improved at least a fair amount as measured according to the Hirabayashi et al.[9] method. Therefore, we strongly support surgical treatment for patients with cervical OPLL who have symptomatic myelopathy, even in those with no radiological evidence of severe disease.

It is well known that surgical outcomes for thoracic myelopathy related to OPLL are worse than those for cervical OPLL.[33,49] Authors of many studies have reviewed the indications and results of the different approaches without establishing a definitive standard of treatment.[6] Several factors increase the difficulty and operative risks for patients with thoracic OPLL. First, the natural kyphosis at this spinal level decreases the effectiveness of decompressive laminectomy because posterior shifting of the spinal cord is restricted. Second, the spinal cord is vulnerable at the site of compression because of poor vascularity. Third, the anterior approach is limited by the presence of the rib cage.[47]

Removal of the posterior longitudinal ligament may be the most effective method of relieving pressure on the spinal cord in patients with thoracic myelopathy related to OPLL. Treatment via a posterior approach with extensive decompression could induce postoperative kyphosis, eliminating the effectiveness of the procedure.[48] To avoid inducing kyphosis, surgeons began using laminoplasty or fusion with a bone graft in combination with instrumentation. However, the outcomes associated with this technique are not ideal, because of the limiting posterior shift of the spinal cord.[46,47]

In 1990, Tomita et al.[41] described a new technique for circumspinal decompression that included the safe removal of OPLL plaque. In 2001, this technique was improved by the introduction of the concept of dekyphosis stabilization.[18] It is now widely accepted that the ossified mass must be completely removed to achieve a full recovery from OPLL-derived myelopathy and that the posterolateral approach is an effective and safe procedure for achieving an optimal outcome.[48] In our single case experience, laminectomy and fusion from T-6 to T-10 was performed without complications. Although the mass was not removed, the patient's postoperative improvement was excellent at his last follow-up visit at 57 months, with no evidence of disease progression. Unfortunately, 1 case precludes a comparative analysis among surgical procedures and clinical outcomes. However, this patient did recover almost fully from the disease, an outcome that supports other reports of promising results following the surgical treatment of thoracic OPLL.

Conclusions

Our findings provide further evidence of the prevalence of a pathology that was once called "the Japanese disease" among whites. As the results of our study show, the demographic and radiographic factors of OPLL are similar in both Asian and white populations, and as in the Asian population, the etiology and potential risk factors in the white population remain unknown. The most appropriate surgical treatment for OPLL remains elusive. We could identify no specific indications for selecting a particular treatment option. All techniques were associated with promising outcomes regardless of the number of involved diseased levels or the radiographic or pathological presentation. The clinical improvement rate was significant, and no surgical complications occurred; therefore, surgery appears to be a safe and effective treatment for OPLL at any spinal level.

Disclosure

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

Author contributions to the study and manuscript preparation include the following. Acquisition of data: Kalb, Martirosyan, Kalani. Analysis and interpretation of data: Perez-Orribo. Drafting the article: Kalb. Reviewed final version of the manuscript and approved it for submission: Theodore. Statistical analysis: Kalb. Administrative/technical/material support: Kalb, Martirosyan. Study supervision: Theodore, Kalani.

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Address correspondence to: Nicholas Theodore, M.D., Division of Neurological Surgery, Barrow Neurological Institute, c/o Neuroscience Publications, 350 West Thomas Road, Phoenix, Arizona 85013. email: nicholas.theodore@bnaneuro.net.